

**MARK ONSLOW**

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**STUTTERING  
AND ITS  
CLINICAL  
MANAGEMENT  
TWELVE LECTURES**

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**OCTOBER  
2024**



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Jerry Siegel  
1932–2014

## **PREFACE**

These lectures are intended as a reference for students of speech-language pathology who are learning to provide clinical services for those who stutter. Regardless, they may be of interest to a broader audience within the speech-language pathology discipline. This text is freely available from the website of the Australian Stuttering Research Centre.<sup>†</sup> It is updated regularly to include newly published research findings and to take account of feedback from users. The year and month of the last update appear on the cover and at the top right of alternate pages.

The lectures constitute a personal view about the basic reference material that students of speech-language pathology need to support adequate clinical services for stuttering. That personal view involves judgements about research publications that do not need to be included in such basic reference material. Much of this introductory course is straightforward. However, much of it is complex material that, at present, leaves more questions than answers. Even so, all of it, in my view, is directly or indirectly applicable to clinical practice.

The writing of this material would not have been possible without the bristling intellectual climate in which I have thrived for past decades. Many have influenced the present work, but most directly I am indebted to Ann Packman, Sue O'Brian, Ross Menzies, and Robyn Lowe. I am particularly indebted to Robyn Lowe for assistance with the writing and to Damien Liu-Brennan for scientific copy editing. Sabine Van Eerdenbrugh assisted with helpful comments on a recent version.

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## LECTURE ONE: BASIC INFORMATION

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### TERMINOLOGY

#### Stuttering

Worldwide, the term *stuttering* is used most often. The term *stammering* is sometimes used in the United Kingdom and Ireland. However, most publications use the term *stuttering*. Several other terms are used in publications, and the use of these terms involves several issues that have been documented in a seminal textbook about stuttering,<sup>1</sup> and by the American Speech-Language-Hearing Association.<sup>2</sup> Some authors recently suggested that researchers and clinicians simply use the term *stuttering*.<sup>3</sup> Issues and potential confusion associated with other terms are now outlined.

#### Dysfluency and dysfluent

The terms *dysfluency* and *dysfluent* are often used to refer to stuttering. However, an issue with these terms is that dictionary definitions of the prefix *-dys* indicate that it means “undesirable” or “abnormal.” As discussed shortly, such implied value judgments are not consistent with some current thinking about stuttering.

#### Disfluency and disfluent

These terms are used to refer to stuttering, but not exclusively. For example, they have been applied to autism<sup>4</sup> and Down Syndrome.<sup>5</sup> They are potentially confusing because they are used also to refer to the usual hesitations and repetitions that can be a part of typical disruptions to the flow of speech that occur with every speaker. Examples of typical disfluency are “well, um, ... gosh, I don’t know,” and “er, I think, perhaps, um, I will get back to you.” The term *typical disfluency* occurs often during these lectures, particularly when referring to the typically developing speech of early childhood.

#### Fluency and fluent

These terms are commonly used in connection with stuttering. The 2013 edition of the Diagnostic and Statistical Manual of Mental Disorders,<sup>6</sup> generally known as the DSM-5, introduced the term *childhood-onset fluency disorder*, and presents it interchangeably with the term *stuttering*. However, arguments have been presented<sup>3</sup> that the terms *fluency* and *fluent* refer to desirable or ideal states. Again, as discussed shortly, such implied value judgments are not consistent with some current thinking about stuttering. Additionally, those authors argue<sup>3</sup> that the terms are misleading because they imply that stuttering involves binary states of either fluent or not fluent, when in fact it may exist on a continuum (as discussed during Lecture Two).

Another limitation of the terms *fluency* and *fluent* is that, as they are commonly used in English, they do not specifically refer to anything about stuttering. They refer to a range of things about the flow of speech,<sup>7</sup> such as rate, prosody, continuity, and smoothness. The term *fluent*, for example, can be used to mean someone speaking a second language proficiently. The same semantic issues pertain to the opposites of these terms, *nonfluency* and *nonfluent*, that are sometimes used for stuttering.

#### Identity-first and person-first terms: Neurodiversity and ableism

##### *Identity-first and person-first terms*

Historically, those who stutter have been referred to with identity-first labels: *stutterer* and *stutterers*. Person-first terminology is a different approach, intended to avoid labelling someone with a condition. Instead, the reference shifts to someone who has a disorder. Accordingly, terms that have been used in recent decades are *a person who stutters*, *someone who stutters*, or *those who stutter*.

Terminology about stuttering is in a continuous state of change. Recently some authors have noted that those who stutter may prefer to be referred to as *stutterers*.<sup>3,8,9</sup> Indeed, recent publications show

signs that the term is returning to use. Another author used the terms *stutterer* and *person who stutters* interchangeably.<sup>8</sup> The author justifies this because those in the disability community have mixed preferences about terminology, and their views should be considered.<sup>10</sup> Those authors<sup>10</sup> cite the example of those who in the deaf community preferring the term “deaf” rather than “people who are deaf.” The style guide of the American Psychological Association<sup>11</sup> recommends taking account of such preferences when deciding about terminology. Clearly, then, when considering how to refer to those who stutter, clinicians need to take account of the preferences of their clients.<sup>†</sup>

#### *Potential limitations of person-first terms*

For all their possible benefits, there are potential limitations with person-first terms for stuttering. Two research publications<sup>12,13</sup> raise doubt about whether person-first terminology for stuttering alters negative perceptions about it. Additionally, person-first terminology involves longer terms than identity-first terminology, and the value of acronyms to deal with that matter has been questioned.<sup>10</sup> With recent stuttering research, the following acronyms have featured regularly: PWS, PWNS, CWS, CWNS. These represent “person who stutters,” “person who does not stutter,” “child who stutters,” and “child who does not stutter.”

Person-first terms invoke present tense, and this can cause awkward expression when writing with past tense. For example, “the research participants were people who stutter.” At least one publication<sup>14</sup> has declined to use person-first terminology to avoid the wording problems that it causes. Additionally, it might be argued that the semantics of person-first terminology are misleading about the nature of stuttering. The term *people who stutter* might imply that stuttering is something that speakers do when speaking, rather than it being something that happens to them when they speak. The latter is what in fact happens, as will be discussed during Lecture Three.

#### *Neurodiversity and ableism and models of disability*

Neurodiversity is the idea that differences in brain function and cognition are best viewed as natural variations of human biology rather than as pathological conditions that should be repaired as much as possible. Ableism is the direct or indirect discrimination against any kind of physical or mental impairment, based on the notion that it is better to not have such impairment. Ableism is sustained by social stereotypes and stigma. These issues centre on a distinction between the social and the medical model of disability. The social model involves disability as a condition created by an unaccommodating society; the medical model of disability locates the issue within the person who has the condition.

The idea of neurodiversity emerged originally in the context of autism spectrum disorder and other conditions such as learning disability and ADHD,<sup>15</sup> and lately has been applied to stuttering.<sup>16</sup> A discussion about these issues<sup>17</sup> dealt with clinical application of neurodiversity and ableism to pre-school children who stutter. The discussion also raised the critical point that a social and a medical model of stuttering can coexist during research and clinical practice.

### **Observable stuttering moments**

#### *The idea of stuttering moments*

The idea of *stuttering moments* is that, to an observer, stuttering seems to be typical speech except for instances when stuttering can be observed. A series of experiments which concluded during the late 1980s established, overall, that the speech of those who stutter indeed does sound typical even when observable stuttering moments are not occurring.<sup>18</sup>

The idea of stuttering moments appears to have emerged during the early 20<sup>th</sup> Century at the University of Iowa.<sup>19</sup> The idea appears regularly in subsequent research literature, however the first formal statement of it appears to have occurred some 30 years later:

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<sup>†</sup> These lectures use identity-first and person-first terms for stuttering.

the stuttering problem might be approached fruitfully by concentrating on *the moment of stuttering*—that is to say, by dealing with the problem of stuttering as a series of stutterings, by regarding it crucially not as a more or less constant condition, but as intermittent responses. (p. 13)<sup>20</sup>

### *The lived experience of stuttering and observable stuttering moments*

As discussed shortly, those who stutter experience it in many ways. A small part of that experience is observable by others, during what can be referred to as *stuttering moments* or *moments of stuttering*. This is a significant distinction for researchers and clinicians to consider: the difference between observable stuttering moments and the experience of stuttering.<sup>3</sup> The experience of stuttering is continuous rather than being constrained to stuttering moments. A survey of 502 stutterers<sup>21</sup> indicated that, at times when they are not stuttering,

only 54.0% reported that they “speak effortlessly.” Notably, 31.2% of the respondents indicated that they only sometimes speak effortlessly, and 26.3% indicated that they only sometimes feel fluent when not in a moment of stuttering. (p. 4339)

As discussed shortly in the context of defining stuttering, much of the experience of it day to day involves a feeling described as loss of control.<sup>22</sup> And such feelings do not necessarily correspond in time with the observable moments of stuttering.<sup>21</sup> Such feelings may be present without any observable signs of stuttering; 59% of adults who stutter report a “sensation of being stuck,” and 50% report “a loss of control” “either often or always” (p. 4339).<sup>21</sup>

## DEFINING STUTTERING

### There is no single definition of stuttering

Ideally, there would be a single, straightforward definition of stuttering that was accepted by everyone. That ideal would definition make it clear who does and who does not stutter. Unfortunately, though, after a vigorous debate for a decade, beginning during the early 1980s, the search for such a workable and generally agreed definition ground to a halt without resolution.<sup>23</sup>

Still, that debate was productive because it established three approaches to defining stuttering. An important point here is that none of the three definitions can be considered as completely satisfactory. They all have limitations, but they all have some strengths that make them useful in different professional contexts. During the 20<sup>th</sup> Century, three leaders in the field contributed to the development of the definitions described below: Marcel Wingate, Oliver Bloodstein, and William Perkins.

### Objective definitions

#### *The World Health Organization definition*

The most common definitions of stuttering are known as objective definitions. They are also known as behavioural definitions and symptomatic definitions. In 1977, The World Health Organization offered what seems to be the most popular definition to date:

Disorders in the rhythm of speech, in which the individual knows precisely what he wishes to say, but at the time is unable to say it because of an involuntary, repetitive prolongation or cessation of a sound. (p. 202)<sup>24</sup>

A more recent World Health Organization definition has so far attracted less attention:

Speech that is characterized by frequent repetition or prolongation of sounds or syllables or words, or by frequent hesitations or pauses that disrupt the rhythmic flow of speech. It should be classified as a disorder only if its severity is such as to markedly disturb the fluency of speech.<sup>25</sup>

*Wingate's definition*

Another older and commonly cited objective definition of stuttering is Marcel Wingate's.<sup>26</sup>

1. (a) Disruption in the fluency of verbal expression, which is (b) characterized by involuntary, audible or silent, repetitions or prolongations in the utterance of short speech elements, namely: sounds, syllables, and words of one syllable. These disruptions (c) usually occur frequently or are marked in character and (d) are not readily controllable.
2. Sometimes the disruptions are (e) accompanied by accessory activities involving the speech apparatus, related or unrelated body structures, or stereotyped speech utterances. These activities give the appearance of being speech-related struggle.
3. Also, there are not infrequently (f) indications or report of the presence of an emotional state, ranging from a general condition of "excitement" or "tension" to more specific emotions of a negative nature such as fear, embarrassment, irritation, or the like. (p. 488)

*The strength of objective definitions*

Objective definitions of stuttering are useful ways to describe stuttering. Wingate's definition is a comprehensive and compact description of stuttering, and as such it is useful in various professional contexts. For example, clinicians could use it, or variations of it, when describing stuttering to clients and other health professionals.

*Limitations of objective definitions*

Objective definitions of stuttering can be regarded only as descriptions of stuttering, not definitions of stuttering, because they cannot be used to set apart those who do stutter from those who do not. This is because there are no observable speech events that can be recorded with words, and which categorically distinguish between stuttering and typical speech.<sup>27</sup> At some time everyone has typical disfluencies that can be described with the same terms that can be used to describe stuttering moments.

For example, with the World Health Organization definition, it is true that those who stutter will experience "involuntary, repetitive prolongation or cessation of a sound," but anyone will do things from time to time that can be described that way. The same can be said about much of Wingate's definition. For example, everyone has "repetitions" occasionally during speech. This definition has also been criticised because it contains "qualifiers and imprecise terms" (p. 17),<sup>28</sup> such as "readily," "sometimes" and "usually," and because speech dimensions such as "controllable" and "involuntary" are not observable,<sup>29</sup> as should be the case with an objective definition.

**Internal definition***Perkins' definition*

William Perkins' definition of stuttering<sup>30,31</sup> is a "temporary overt or covert loss of control of the ability to move forward fluently in the execution of linguistically formulated speech" (p. 431).<sup>31</sup> This is referred to as an internal definition because "loss of control" refers to a speaker's experience. This contrasts it with the objective, observable features of objective definitions. The experiences of those who stutter commonly align with the internal definition:

Believe me, stuttering is very frustrating. You know what you want to say, and you know how to say it. But when you want to say it, you can't. You're almost jealous of people who can just say whatever they want to say when they want to.<sup>32</sup>

*The strength of the internal definition*

The internal definition of stuttering certainly is a valid one, because stuttering is fundamentally a personalised experience for those affected. The proponents of this definition even conducted an

experiment claiming to verify this.<sup>33</sup> They showed that a speaker could distinguish recordings of real and faked stuttering shortly after producing them, but neither the speaker nor listeners could distinguish them at later times.

Those who stutter commonly report loss of control as part of the experience. As noted earlier, one report<sup>35</sup> was a survey of 502 stutterers, in which 96% reported a “sensation of being stuck” and 94% reported “a loss of control” (p. 4338) during a moment of stuttering. Based on interviews with 13 participants, another report<sup>22</sup> made the important point that a feeling of loss of control at any one moment is not necessarily accompanied by observable speech behaviours that are part of the objective definition of stuttering. The authors also reported an association between loss of control and the anticipation of stuttering, which is discussed later in this lecture.

The authors of one report<sup>22</sup> state a fundamental clinical principle:

Aligning the definition of stuttering behavior with the experiences of individuals who stutter has significant clinical implications for improving diagnostic and therapeutic outcomes. (pp. 1190)

And indeed, clinicians rely on internal definition of stuttering during routine clinical measurement of stuttering severity. As will be discussed in Lecture Four, it is essential to obtain client reports of how severe their stuttering is. When clients give you that information they are, in effect, drawing on an internal definition of stuttering. If a client says that stuttering is not present, and has not been present for a significant period, that is important clinical information because of its validity. Internal definition of stuttering can be useful during research about it. For example, in one report,<sup>34</sup> children were asked if they thought that they stuttered as part of determining whether they had recovered from stuttering.

Another reason why internal definition of stuttering is valid is that it reflects what clinicians want to achieve for clients with treatment: a change of the experience of stuttering, and a positive shift of how they feel it affects them. The obvious validity of the internal definition was shown with a report<sup>35</sup> of 430 adult stutterers who were surveyed about how they thought stuttering should be defined. The researchers concluded that:

To adults who stutter, the term *stuttering* signifies a constellation of experiences beyond the observable speech disfluency behaviors that are typically defined as stuttering by listeners. Participants reported that the moment of stuttering often begins with a sensation of anticipation, feeling stuck, or losing control. (p. 4356)<sup>35</sup>

#### *Limitations of the internal definition*

It has been argued that the internal definition is more a statement about the nature of stuttering than a definition.<sup>36</sup> Also, this definition has in common with objective definitions that it fails to distinguish between stuttering and usual speech. Probably, all speakers would report that, at some time, they lose control of their speech. Another issue is that clinicians cannot directly observe “loss of control” because it is an experience, not a behaviour.

### **Perceptual definition**

#### *Bloodstein’s definition*

Oliver Bloodstein’s definition<sup>37</sup> is that stuttering is “whatever is perceived as stuttering by a reliable observer who has relatively good agreement with others” (pp. 9–10). In other words, a clinician who has consensus with a community of experienced speech-language pathology observers determines whether stuttering is present or whether it is not.

#### *Strengths of perceptual definition*

An advantage of perceptual definition is that, if the required consensus exists, it is procedurally simple and clinically workable. When parents bring children who have just begun to stutter to the clinic, they are reporting their perception that stuttering is present. As discussed shortly, there is reason to believe that clinicians generally agree with parents in such cases. So, it is arguable that parents of children

who stutter are reliable observers who have “relatively good agreement with others,” and so they are using a perceptual definition of stuttering.

#### *Limitations of perceptual definition*

Bloodstein’s perceptual stuttering definition is not clear about what constitutes a “reliable” observer and “relatively good agreement with others.”<sup>29</sup> Indeed, a stuttering definition that relies on clinical judgement that is consistent with a clinical community raises the question of how junior clinicians might attain such consistent judgements. The answer to that is conceptually simple; senior clinicians can mentor junior clinicians about what are appropriate judgements. Even so, it may be that different clinical communities, such as those in various countries, may differ in perceptions about what stuttering is and what it is not. There is some evidence that this may occur.<sup>38</sup>

## DESCRIBING STUTTERING MOMENTS

### Taxonomies

Wendell Johnson<sup>†</sup> developed the first system for classification of stuttering moments.<sup>39</sup> This taxonomy was developed specifically for stuttering during early childhood, and included eight terms: *word repetition, sound/syllable repetition, phrase repetition, incomplete phrase, interjection, revision, broken word, and prolongation*. There have been several variants of this initial taxonomy.<sup>40,41</sup> The better-known terms that were added to Johnson’s original taxonomy are *disrhythmic phonation, block, blockage, and tense pause*. All these taxonomies deal with stuttering during early childhood, except for one.<sup>42</sup> Presumably, this is because of the profound historical influence on the field of two theoretical perspectives about early childhood stuttering, which are reviewed during Lectures Two and Three: the Diagnosogenic Theory and the Continuity Hypothesis.

### Unambiguous stuttering moments

The term *unambiguous stuttering moments* refers to moments during speech that, to an observer, are clearly stuttering and not typical disfluency. When observing the speech of someone who stutters, a clinician sometimes may be unsure whether a particular speech event is a stuttering moment or a typical disfluency—particularly with young children. During clinical practice, this is not normally an issue. However, sometimes after successful treatment of young children, parents need guidance with being certain of the distinction between a stuttering moment and a typical disfluency.

### A taxonomy: The Lidcombe Behavioural Data Language

#### *Overview*

The following method<sup>40</sup> to describe unambiguous stuttering moments arguably has some advantages. It was developed for use with stuttering clients of all ages, and it describes speech behaviours only; it contains no reference to anything that cannot be observed. Additionally, it appears that with some clinical experience it can be used reliably.<sup>43</sup> It is known as the Lidcombe Behavioural Data Language.

This taxonomy presents stuttering behaviours in three prime categories: *repeated movements, fixed postures, and superfluous behaviours*. There is nothing new about these terms. Variations of them have been used for decades: for example, *repetitions, prolongations, and accessory features*.<sup>26</sup> A useful insight into the speech behaviours of stuttering is available with a video of real-time magnetic resonance imaging.<sup>44</sup>

#### *Repeated movements*

Commonly, clinicians refer to these as repetitions. There are three different types of *repeated movements*. The first is *syllable repetition*.

<sup>†</sup> Wendell Johnson, who was located at the University of Iowa, arguably was the most influential figure in the history of stuttering research and theory.



Syllable repetition is straightforward, being a repeated movement of what sounds like an entire syllable. For example, “when-when-when-when,” “if-if-if-if-if,” and “not-not-not-not.”

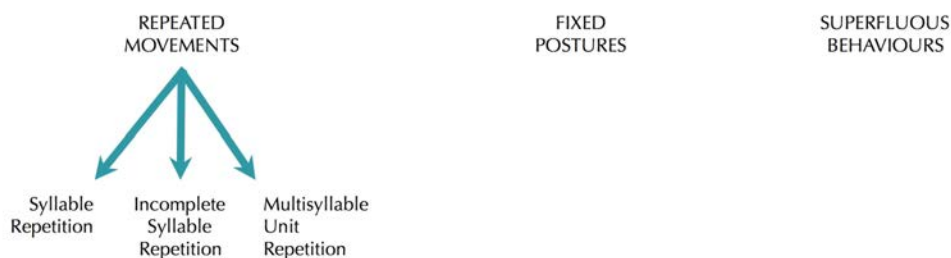
Not all syllable repetitions are repetitions of entire syllables. Some of them are repetitions of parts of syllables, which are termed *incomplete syllable repetition*, meaning that the speaker did not repeat an entire syllable, but part of one.



Some of the distinctions between a syllable repetition and an incomplete syllable repetition may require careful listening to identify. For example, “can-can-can-can” is a repetition of the entire syllable, with all its phonemes. But with “ca-ca-ca-ca-can” the speaker has produced only the first two phonemes of the syllable before eventually saying the entire syllable. In which case it is an incomplete syllable repetition.

Returning to the example of the syllable repetition “not-not-not-not,” if “no-no-no-not” was heard, it would be an incomplete syllable repetition. Also, a syllable repetition might be “I-I-I-I-I.” At first it might seem that this could only be a syllable repetition, but again, careful listening may be needed. The word “I” is a diphthong in most spoken English and the speaker might not complete the two vowel-like parts of this, and instead say something like “uh-uh-uh-uh-I” while attempting to say “I.” In which case, it would be an incomplete syllable repetition.

Repeated movements can also involve more than one syllable, in which case the term *multisyllable unit repetition* is used.

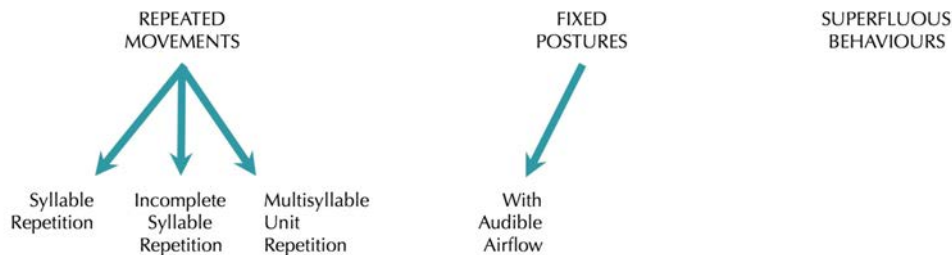


Examples of multisyllable unit repetition are “I was-I was-I was-I was hoping,” “I think that-I think that-I think that-I think that,” and “then-I then-I then-I.”

### Fixed postures

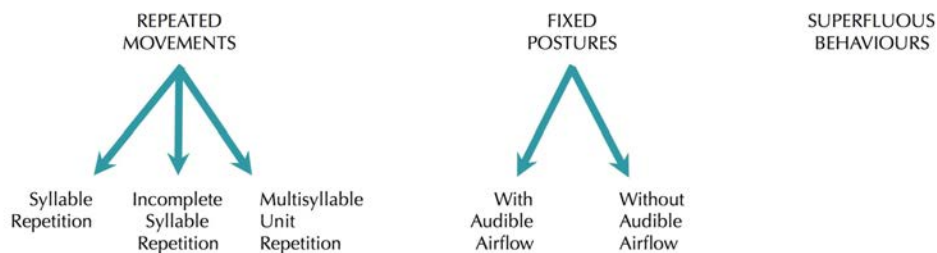
*Fixed postures* are in a sense the opposite kind of stuttering behaviour to repeated movements because they are not an atypical movement but an absence of typical movement. During fixed postures what normally moves during speech—mostly mouth, jaw, and lips—stops moving. It can stop moving for a period so short that skilled listening is necessary to detect it. It is far more obvious when fixed postures happen for a long period of several seconds. In severe cases, fixed postures can stop speech for half a minute, which of course seriously disrupts communication.

The first category of fixed postures is *with audible airflow*.



There are many kinds of airflow that can be audible. These include articulatory and laryngeal fricative noises and, more commonly, phonation. Clinicians often refer to fixed postures with audible airflow as “prolongations,” because that is exactly how they sound: as if the speaker is prolonging a sound.

The second category of fixed postures is *without audible airflow*.



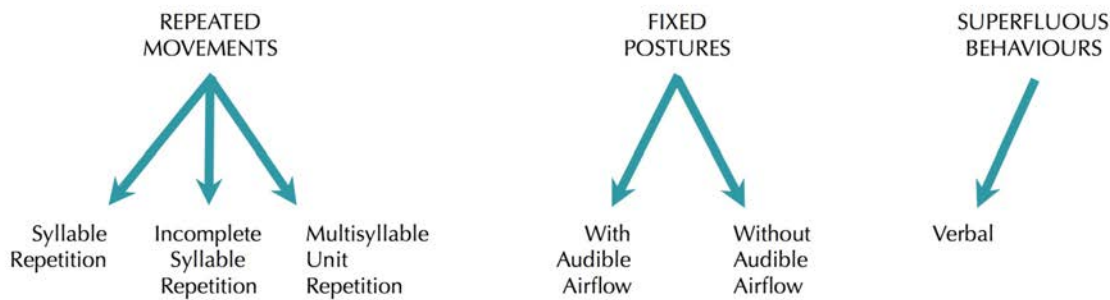
During these no airflow is audible. But it is necessary to listen carefully to be sure that there really is nothing audible. Sometimes the airflow can be barely audible. Clinicians often refer to fixed postures without audible airflow as “blocks,” because they give the impression that something is blocking speech.

### Superfluous behaviours

The final category of stuttering moments is *superfluous behaviours*. These are redundant to the intended meaning of the utterance as it normally would be spoken, hence, the term “superfluous.” These are often the most socially distracting of the observable behaviours of stuttering.

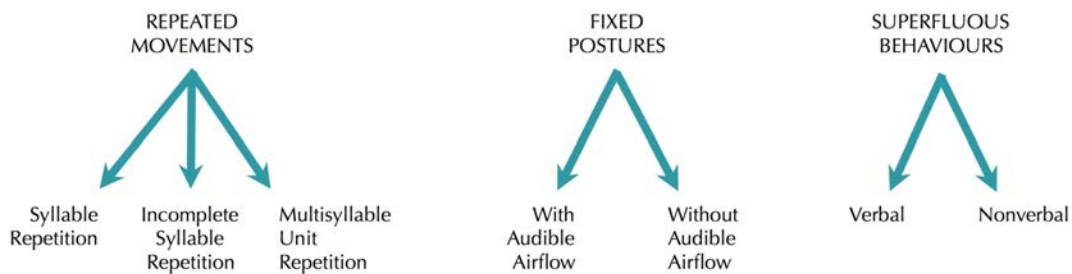
The first kind of superfluous behaviours is *verbal*.





It can be a challenge to identify some verbal superfluous behaviours because it is not clear whether they are redundant to the intended utterance. Johnson's taxonomy refers to them as interjections,<sup>39</sup> which is a term that assists with understanding how they can sound. An example would be "oh well-oh well-well-um-um."

The other kind of superfluous behaviours is *nonverbal*.

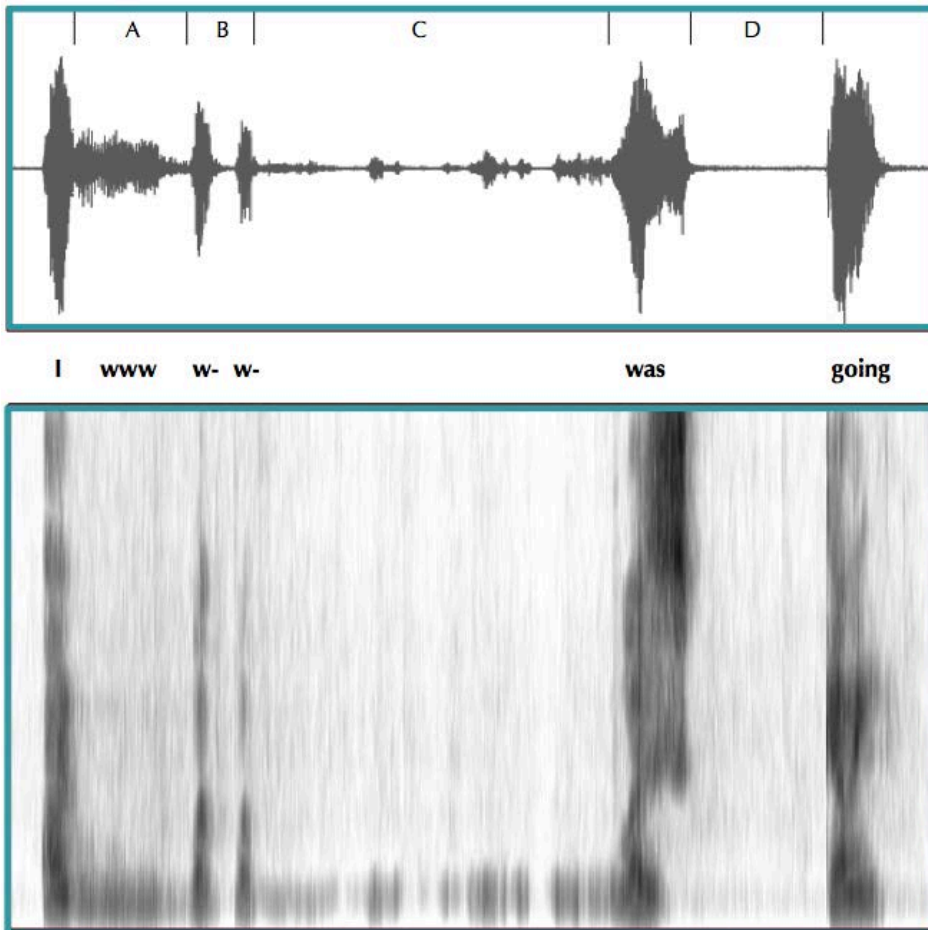


Nonverbal superfluous behaviours are easy to identify because they are obviously redundant to the intended meaning of the utterance. They include compressed lips, open mouth, breath holding, blinking, nostril dilating, eyebrow raising, grimacing, facial, head, and torso movements, inspiratory airflow, grunts and other inappropriate noises, and unusual fluctuations of pitch and loudness. Stuttering is idiosyncratic; it is rare to see two people whose stuttering looks identical, and nonverbal superfluous behaviours are the most idiosyncratic features of it.

### Stuttering behaviours combine in one stuttering moment

The seven stuttering behaviours described with this taxonomy, or with any taxonomy, are not mutually exclusive.<sup>27</sup> One, many, or even all the seven stuttering behaviours, can be present during one stuttering moment.<sup>45</sup> In fact, it is rare for a stuttering behaviour to have only one of the seven stuttering behaviours by itself. For example, a stuttering moment that is a repeated movement could be a syllable repetition and an incomplete syllable repetition at the same time. Such a stuttering moment might sound like "ca-ca-ca-can-can-can." Or a repeated movement could be a syllable repetition, an incomplete syllable repetition, and a multisyllable unit repetition all at once. That might sound something like "ca-ca-ca-can-can-can-I-can I-can I."

The waveform (top panel) and spectrogram (bottom panel) in the following figure show a stuttering moment on "was" that is a fixed posture with audible airflow (Segments A and C) and incomplete syllable repetitions (Segment B). Subsequently, there is a fixed posture without audible airflow during another stuttering moment prior to the word "going" (Segment D).



A study of 3,100 stuttering moments from 147 adolescents and adults<sup>46</sup> reported that around half of the stuttering moments contained a repeated movement, around half contained a superfluous behavior, and around two thirds contained a fixed posture. The most common combination of stuttering moments was fixed postures and superfluous behaviours, and the least commonly occurring combination contained superfluous behaviours only. For 18% of the stuttering moments, repeated movements, fixed postures, and superfluous behaviors occurred together.

### Some clinical applications of describing stuttering

#### *Communicating with other clinicians*

When writing about a client to another clinician, it may be preferable to incorporate formal terminology, such as in this example:

Most of Mr Williams' stuttered speech contained fixed postures with audible airflow, with his jaw almost shut and airflow comprising alveolar fricatives. Most of these lasted more than 1 second, with several of them lasting more than 10 seconds. During these fixed postures he had extraneous nonverbal behaviours, often grimacing with his eyes closed, brow furrowed, and head tilted downwards and to the left. None of Mr Williams' stuttering moments involved repeated movements.

#### *Communicating with other professionals*

With reports about clients to other professionals, such as a teacher or doctor, more general terminology may be preferable to such discipline-specific terminology:

Most of Mr Williams' stuttered speech contained speech blocks with audible airflow, with his jaw almost shut. Most of these speech blocks lasted more than 1 second, with several of them lasting more than 10 seconds. During these blocks he had nonverbal behaviours, typically grimacing with his eyes closed, brow furrowed, and head tilted downwards.

## THE DISTRIBUTION OF STUTTERING MOMENTS

### The influence of spoken language

#### *Initial word consonants*

Stuttering moments do not occur randomly during speech. Early during the last century, seminal research from the University of Iowa<sup>47</sup> showed that their occurrence follows rules to a considerable extent. Stuttering was shown to occur more commonly on consonants than on vowels, with most stuttering—more than 90%—occurring on the initial sounds of words. That finding has been replicated<sup>†</sup> many times,<sup>48</sup> although one failure to replicate has occurred.<sup>49</sup> The effect has been shown to occur with Japanese children who stutter,<sup>50</sup> with a mean age of 5 years 9 months, and with Turkish children 6–16 years old.<sup>51</sup>

#### *First word of an utterance*

In addition to occurring commonly on the first sound in a word, stuttering moments occur commonly on the first word of an utterance,<sup>52,53,54,55</sup> although, again, a failure to replicate has occurred.<sup>56</sup> The effect has also been reported for the first word of clauses.<sup>57</sup> This effect has been found with Turkish stuttering children 6–16 years old.<sup>51</sup>

#### *Rare at the end of words*

It also appears that sometimes—but rarely—stuttering can occur with repeated movements at the end of words.<sup>58</sup> This is sometimes referred to as word-final disfluency. However, queries have been raised that such repeated movements may not be connected with stuttering. A treatment report of 20 children 5–11 years with word-final disfluency<sup>59</sup> indicated that none were diagnosed with stuttering (five were diagnosed with autism). One report<sup>60</sup> documented behaviours other than repeated movements at the end of words, but noted that they are difficult to identify perceptually.

#### *“Difficult” sounds*

Clients commonly report that certain sounds are “difficult” because stuttering is likely to occur with them. The seminal report, referred to previously,<sup>47</sup> showed individuality among those who stutter for sounds that are stuttered more often and considered to be “difficult.” Another report from the same laboratory<sup>61</sup> verified this finding, by showing that there is no general rule about which sounds are difficult for those who stutter.

#### *Content words*

Another early report from the University of Iowa<sup>62</sup> indicated that traditional grammar influenced the occurrence of stuttering. Words with heavy semantic content, such as nouns, verbs, and adjectives, are stuttered more often than words with little semantic content, such as articles, conjunctions, and interjections. Or, to say it another way, stuttering is more likely to occur on content words than on function words. This finding has been replicated many times, and there is some suggestion that the effect may be language specific, with a report that it does not occur in Arabic.<sup>63</sup> However, it has been reported to occur with Persian<sup>64</sup> and Nepali speakers who stutter,<sup>65</sup> and with Turkish children who stutter.<sup>51</sup> One report<sup>66</sup> suggested that with bilingual speakers the effect might be present in the first language but not the second.

Intriguingly, there are replicated findings that with children the effect is reversed. There are reports of

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<sup>†</sup> It is a general rule that a research finding is not particularly believable unless it has been reported by independent researchers.

more stuttering occurring on function words than on content words for English,<sup>67,68,69,70</sup> German,<sup>71,72</sup> Spanish,<sup>73</sup> and Spanish-English bilingual<sup>74</sup> children. A report with Korean-speaking and English-speaking 3–7-year-olds<sup>75</sup> found that this effect was reversed for the Korean-speaking children, with them stuttering more on content words than function words. Two reports<sup>73,87</sup> have reported such a finding with studies of adults compared to children. This effect is of interest when attempting to understand the cause of stuttering, as will be discussed in Lecture Three.

### *Stressed syllables*

Some reports have found that stressed syllables are stuttered more often than unstressed syllables,<sup>48,76</sup> although others have failed to find such an effect.<sup>77,78,79,80</sup> A report has extended such research to lexical tone with 20 Mandarin speaking Taiwanese children with early stuttering, with a mean age of 4 years 9 months.<sup>81</sup> Results showed that “stuttering-like disfluencies” (see Lecture Four) were around twice as likely to be associated with syllables carrying Tone 3 or Tone 4 compared to syllables carrying Tone 1 or Tone 2. The authors plausibly speculated that results “may be attributed to the increased level of speech motor demand underlying rapid F0 [*fundamental frequency*] change both within and across syllables” (p. 115).<sup>81</sup> This explanation might apply also with findings in English of more stuttering on initial word consonants and stressed syllables. However, a report about stuttering in Cantonese<sup>82</sup> found no differences for stuttering moments across its six tones. A study of Japanese child stutterers,<sup>50</sup> with a mean age of 5 years 9 months, reported no differences between “heavy and light syllables.”

### *Utterance length and grammatical complexity*

Increased utterance length<sup>†</sup> is associated with increased syntactic complexity, and has been associated with increased stuttering.<sup>77,83,84,56</sup> These findings have been replicated many times with children,<sup>85</sup> although it is probably fair to say that the findings are not as marked and consistent as with adults. Consistent with these findings are reports that long words (measured with syllables or letters) are stuttered more often than short words.<sup>86,87</sup> Some authors have noted<sup>88</sup> that increased utterance length is associated with decontextualised language that is outside the “here and now,” comprising “extended, abstract discourse that is removed from the physical context of an interaction” (p. 2657). They reported that, compared to controls, decontextualised utterances of pre-school children were more likely to be stuttered. A report<sup>89</sup> linked the effect of utterance length on stuttering to a measure of speech motor function (lip aperture variability) with a group of 7–12 year old child stutterers and a control group. The child stutterers showed more lip aperture variability than controls as utterance duration increased. This is consistent with current perspectives of stuttering as an issue with atypical neural processing of speech, as will be discussed during Lecture Three.

### *Clustering*

Clustering is another way that stuttering moments occur non-randomly during speech. Clustering is the occurrence of a series of stuttering moments shortly after each other. This has been reported many times for early childhood stuttering,<sup>90</sup> and with adults.<sup>91,57</sup>

### *How predictable are stuttering moments?*

An early report<sup>87</sup> indicated that 95% of stuttering moments could be accounted for by “initial sound, grammatical function, sentence position and word length” (p. 183). A later source<sup>86</sup> was consistent with that finding, reporting that 95% of stuttering moments can be accounted for by the word initial phoneme, grammatical class, word length, and word position in the utterance. In other words, most stuttering moments occur according to rules, but it is not possible to fully account for the occurrence of every stuttering moment. A study of the conversations of 35 adults who stuttered,<sup>92</sup> using a publicly available data bank, explored several predictors of stuttering moments: “initial phoneme,” “grammatical function,” “word length,” “word position within a sentence,” “word frequency,” and “neighborhood density” (surrounding words that are phonologically similar) (p. 6). Regression analyses

<sup>†</sup> Utterance length is usually measured with words, syllables, or morphemes.

indicated that these six factors could predict stuttering moments with 64% accuracy.

### Adaptation, consistency, and adjacency

#### *The adaptation effect*

After around five readings of the same passage, stuttering decreases on average by half. This so-called *adaptation effect* was a much researched aspect of stuttering during the last century. Why the adaptation effect occurs is not clear. There are data to suggest that it is caused by subtle changes in speech motor function that occur over successive readings,<sup>93</sup> and there are data to suggest the opposite.<sup>94</sup> There is also some evidence that motor learning may explain it,<sup>95</sup> and that motor learning may interact with the language spoken.<sup>96</sup> The motor learning explanation is bolstered by evidence that the adaptation effect seems to occur with the “stuttering-like disfluencies” (see Lecture Four) of Parkinson’s disease.<sup>97</sup> The effect might also be explained by anxiety about speaking systematically reducing after several readings.<sup>98,99</sup> Adding to this perplexity, there is a report that fluently spoken syllables just prior to a stuttered syllable are lengthened, but there is no change in that pattern over successive readings.<sup>100</sup> In summary, it is possible, or even likely, that an interaction between speech motor function and anxiety factors is involved with the adaptation effect.

#### *The consistency effect and the adjacency effect*

It is likely that the *consistency effect* and the *adjacency effect* are involved with the adaptation effect. The consistency effect is that stuttering tends to occur on the same words during repeated readings of a passage.<sup>101,102</sup> The adjacency effect is when stuttered words are removed from a passage and it is read again, and stuttering tends to occur on words located near the removed ones.<sup>103,104</sup>

## HOW STUTTERING AFFECTS PEOPLE

### Treatment seeking

Those affected by stuttering commonly seek treatment. An Australian survey study<sup>105</sup> of 852 adult stutterers, with a mean age of 49 years, indicated that 72% of them were assessed by a speech-language pathologist, and 73% received treatment from a speech-language pathologist.<sup>†</sup> As with many health issues, early treatment is a desirable option for stuttering, as discussed during Lectures Six and Seven. For 135 children in that survey, with a mean age of 11 years, parents reported that 95% received assessment and 92% received treatment from speech-language pathologists. This section describes many reasons why those who stutter might seek clinical services. Stuttering affects speech, quality of life, educational and occupational attainment, and mental health.

### Speech impact

#### *Reduced verbal output*

A self-evident but much overlooked impact of stuttering is reduced verbal output. This occurs because the speech behaviours described earlier in this lecture are time consuming. Those who stutter appear not to say as much as their peers within a given time, or take longer to say it, or a combination of both. According to an early study of the matter,<sup>106</sup> when given a spontaneous speaking task, those who stutter say, on average, around one third less than those who do not stutter. A more recent publication<sup>107</sup> replicated that finding. A group of control speakers had a mean of 867 words spoken in 5 minutes compared to a mean of 584 words for a stuttering group, which is one third less.

With severe stuttering, speech rate can be below 50 syllables per minute, which is speech output less

<sup>†</sup> Throughout these lectures the North American term *speech-language pathologist* is used. However, those who are professionally qualified to provide clinical services for stuttering are referred to with several names. Examples are speech pathologist (Australia), speech and language therapist (United Kingdom and Japan), Logopäd (Germany), orthophoniste (France), logopædagog (Denmark) and logopedist (Netherlands).

than a quarter of peers. So, a person severely affected for a lifetime may say only a quarter of what is possible, or may take four times as long as others to say what is intended.

#### *Variable stuttering severity*

Stuttering severity is notoriously variable; "...you have some good days and some not so good days."<sup>108</sup> A survey of 204 adults,<sup>109</sup> mostly from the United States, indicated that 97% of them experienced variability of stuttering severity. Findings indicated this to be the most frustrating aspect of stuttering for them.

Stuttering severity is likely to vary with differing audience sizes and types,<sup>110,111,112</sup> generally with more stuttering as audiences become larger. Stuttering severity varies also across different everyday situations.<sup>113</sup> It seems that there will be more stuttering when speaking to people than when speaking alone, as discussed at the end of this lecture. Experiments that have involved repeated measures of participants in the same speaking situation have shown clinically significant stuttering variability in that same situation.<sup>114,115,116</sup> A study of six participants over five clinic visits spanning 2 weeks<sup>117</sup> showed that in two cases stuttering severity was four or five times greater on some visits than others. There is a report<sup>118</sup> that stuttering changes even with different postures, with more stuttering when lying down than when sitting.

Statistical process control charts are a method of studying variation, and this method has been applied to stuttering.<sup>119</sup> The stuttering severity of 10 adults was studied during a single day. Results showed that all 10 participants showed predictable variation around their mean severity. However, five of the participants had stuttering severity that was unpredictable across the day, and suggestive of an "out of control system," with severity scores more than three standard deviations from their means during the day.

#### *Word avoidance*

Those who stutter may attempt to limit the impact of their stuttering on daily life by avoiding words.<sup>120,121,122</sup> Scanning ahead for words that are difficult, and avoiding them with circumlocutions, is a common strategy, as is word substitution:

I am a thesaurus. I know a synonym for every word that I can think of. So if I'm having difficulty saying it, I can immediately come up with another word that I can substitute for it.<sup>32</sup>

However, there are some words that cannot be avoided, and this can negatively impact daily life. Commonly, those who stutter have difficulty saying their names:

I was in a shop a couple of years ago and the sales rep asked me for my name. I tried to get it out but I was having problems. He asked me on the spur of the moment, which I think is even a bit more difficult sometimes. I couldn't say my name. He said to me, "Don't you know your own name?" And I was really affected by that.

And I've known people who stutter who changed their names, or they use a nickname. And that's okay for a while. But then once that nickname becomes a part of their identity, they start to stutter on that name as well.<sup>32</sup>

Other common examples of words that cannot be avoided are telephone numbers, addresses, destinations on public transport, and food items:

Even ordering food over the phone. That seems to be all right. But when I order delivery and I have to give my address, I don't know why that's the issue, when I have to give my address. Because you ... can't substitute a "two" for a "five" or a "four" for a "seven." So I don't do delivery.

I used to love egg and lettuce sandwiches. Unfortunately, egg was hard to say back then before I had treatment or if my speech wasn't particularly good on that day. So chicken, lettuce and mayonnaise soon became my favourite sandwich.<sup>32</sup>

You are at your favorite bakery, craving a pain au chocolat. As you attempt to order, your jaw, lips, and tongue will not move and no air flows through your voice box—you struggle to get the speech sound out, but however much you try, you are frozen in place and there is no sound. You cannot answer the clerk's friendly greeting, and to your dismay you can now see that she looks uncomfortable, unsure of how to react. Looking to escape the awkward situation, you end up ordering something you do not want at all, only because it has a name that you can say without trouble. (p. 1)<sup>123</sup>

### *Grammatical constraints*

There have been reports that those who stutter have restricted use of grammar.<sup>107,124</sup> The latter of these papers reported that, compared to controls, those who stutter spoke with fewer clauses per utterance and fewer elaborate clause constructions. Also, the stuttering group used less modality than nonstuttering peers. The term modality, in systemic functional linguistics, refers to “linguistic resources to express opinions, attitudes, and politeness, and therefore potentially engage with conversation partners” (p. 481).<sup>107</sup> A particularly noticeable reduction of modality occurred with interpersonal metaphors, indicating that the stuttering participants were less inclined to project opinions with clauses such as “I believe ...” and “I think ... .” These results, with traditional grammar and systemic functional linguistics, were replicated in a more recent report,<sup>125</sup> where the authors concluded that those who stutter have “a reduced openness to interpersonal engagement within communication exchanges” (p. 536).

A follow-up report on the participants in the latter study after speech treatment<sup>126</sup> indicated some improvement in flexible language use related to interpersonal engagement, but not a complete resolution of the issues. A 12-month follow-up<sup>127</sup> showed these treatment gains to be maintained, with evidence of continued improvement. There is also a suggestion from a study<sup>128</sup> of eight 7-year-old child stutterers and eight control children that such problems may begin early in life. There has since been initial work to develop a questionnaire assessment of these pragmatic language functions.<sup>129</sup>

### **Quality of life impact**

The importance of stuttering and quality of life was highlighted by an issue of the *Journal of Fluency Disorders* being devoted to the topic.<sup>130,131,132,133</sup> The quality of life impairment that stuttering can cause is well demonstrated in a film depicting the life of King George VI.<sup>134</sup> The United States President Joe Biden stuttered severely as a child and remains affected as an adult.<sup>135</sup> He described stuttering as “the single most defining thing in my life”<sup>136</sup>

Two studies<sup>137,138</sup> used a medically oriented quality of life instrument to show that stuttering participants had poorer quality of life compared to controls. Two reports with adults indicated health economics utility scores from 0–1, spanning death (0) to perfect health (1): 0.83<sup>139</sup> and 0.71.<sup>140</sup> A systematic review of medical quality of life utility scores for patients<sup>141</sup> suggests that these scores are in the range of those for chronic health conditions: 0.83 for diabetes mellitus, 0.77 for cardiovascular disease, 0.75 for cancer, 0.84 for HIV, and 0.70 for chronic kidney disease. Presumably this is because, in contrast to those diseases for the most part, stuttering is present across the lifespan, beginning during the first years of life. Indeed, a study measured quality of life impairment from stuttering early in life, during the pre-school years, with a utility value of 0.88.<sup>142</sup>

These utility scores were noted in a systematic review,<sup>143</sup> and two of the three studies that involved utility scores were considered to have a high risk of bias. The authors of the systematic review concluded that there was limited evidence, using standard utility scores, of quality of life impact from stuttering, and its improvement after treatment. They called for that situation to be remedied urgently so that the quality of life impairment associated with stuttering can be well documented and used to influence governments and other health care funders to provide adequate resources for its health care.

Another study<sup>144</sup> recruited 78 participants, four of whom stuttered, to a “willingness to pay” and “quality adjusted life years” analysis of stuttering. The nonstuttering participants were provided with detailed information about stuttering, and results indicated that participants would pay “with amounts of money equal to two to four times their annual incomes” (p. 309)<sup>144</sup> for a clinical improvement to

mild or “cured” stuttering. Additionally, respondents “equated substantial improvements in severe stuttering with a gain of up to 18 additional years of full-health life” (p. 309).<sup>144</sup> These results were consistent with quality of life impairments measured for serious medical illnesses.

A report has linked the quality of life impairment of stuttering to the lack of speech spontaneity it induces.<sup>145</sup> There is some evidence that stuttering restricts participation in some forms of sport and exercise.<sup>146</sup> There is evidence that teenage girls who stutter are affected more by stuttering than teenage boys.<sup>147</sup>

Bandura’s<sup>148</sup> notion of self-efficacy refers to the extent to which people believe they will be able to achieve things. With reference to those who stutter, a report<sup>149</sup> of 39 adults found a relationship between high levels of self-efficacy and low impact scores on the Overall Assessment of the Speaker’s Experience of Stuttering.<sup>†</sup> The effect was found to occur independent of stuttering severity. For those who stutter, self-efficacy was found to relate to a feeling of spontaneity while speaking.<sup>145</sup>

### Cost of illness

Combined with health utility scores, cost of illness data are part of comprehensive health economics analyses. Cost of illness measures direct and indirect burdens to society, in monetary terms. Direct costs include treatment expenses and lost work hours. A systematic review<sup>143</sup> concluded that the direct cost of stuttering treatment expenses for adults is around US\$1,000 per year, and commonly used early childhood treatments are in the range of US\$4,000–4,500 per year. Indirect costs to society include carer burden, costs to health and education systems, and restricted social functioning because of health issues. The review<sup>143</sup> concluded that the direct and indirect costs associated with stuttering

include impairment, challenges, and distress across many domains throughout life, including income, education, employment, and social functioning. (p. 1)

These topics are covered in the following sections that overview the occupational and educational impact of stuttering, and societal stereotypes and stigma about it.

### Occupational impact

#### *The modern importance of communication and occupation*

During the past century there has been systematic change with how much speech is needed for common occupations, and this has implications for those who stutter. In the United States,<sup>150</sup> 80% of occupations relied on manual skills at the start of the 20<sup>th</sup> Century, with only 20% of occupations relying prominently on communication skills. By the 1950s the proportion of such “white collar” occupations relying on communication skills had increased to 38% and the figure was 62% at the start of this century. For Australia, in 1966, 45% of occupations were “white collar,” rising to 69% in 2011.<sup>151</sup>

#### *Stuttering impairs occupational attainment*

Stuttering affects daily experiences in the workplace,<sup>152,153,154</sup> and can be classified as a major discrimination experience in life.<sup>155</sup> Considering the importance of communication in the workplace, it is not surprising that stuttering has an impact on occupational attainment. A survey that included 713 adult stutterers<sup>105</sup> reported that 55% indicated that stuttering had been a barrier to finding employment, and 66% indicated that it had been a barrier to career progression. Another report<sup>156</sup> showed that 70% of 200 stuttering adults thought that it prevented promotion and 20% declined the challenge of a promotion because of stuttering. Some who stutter even recount employment their employment being terminated because of their stuttering: 7.5% in one report<sup>157</sup> and 5.3% in another.<sup>155</sup> Another report indicated that speech rehabilitation resulted in improved occupational level and promotion prospects.<sup>158</sup>

Employers have reported that those who stutter are less employable or promotable than others.<sup>159</sup> A

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<sup>†</sup> This is a measure of the impact of stuttering, which is discussed in Lecture Four.



study of Turkish employers reported negative stereotypes about stuttering.<sup>160</sup> Members of the public seem to reflect these attitudes.<sup>161,162</sup> A study of questionnaire responses of 20 adults in Jordan<sup>163</sup> showed that the following beliefs were ranked highly from a list of 10: “you face challenges to get a leadership position because of your stuttering” (ranked first); “employers exclude you from getting a job because of your stuttering” (ranked third). These beliefs seem to occur also in Japanese society.<sup>164</sup> A survey study of human resource management students in the United States<sup>165</sup> found that 42 of 43 potential careers were thought by the students to be less suited to someone who stutters than someone who does not. It seems that it often occurs in educational settings for those who stutter to be discouraged from pursuing certain careers, or even to be discouraged from pursuing education.<sup>155</sup>

A study of a large British birth cohort<sup>166</sup>—participants studied from birth—supported these findings by indicating that those who stutter are more likely to have lower socioeconomic occupation status than those who do not. A large United States cohort<sup>167</sup> observed from childhood produced more definitive findings: those who stuttered earned less annually compared to controls, and those who stuttered were more likely to be unemployed than controls. The study reported a particular disadvantage for women who stuttered compared to men who stuttered. A study using another large United States cohort<sup>168</sup> reported that those who had received treatment for their stuttering from a speech-language pathologist had substantively higher income than those who had not received treatment.

On the positive side, a Japanese study<sup>169</sup> endorsed the practice of companies having employment quotas for those with disabilities, stating that “currently, 103 countries provide employment quotas in their national legislation” (p. 2). Twelve adult stutterers, who were hired under such a quota system, were interviewed. Overall, they did not feel discriminated against or report receiving a lower salary than other employees.

## Educational impact

### *The school years*

Some early publications identified education problems for children who stutter during the school years,<sup>170,171</sup> and those results have been replicated in a more recent report.<sup>172</sup> A compelling, large cohort,<sup>173</sup> based on 1988 data from The United States National Health Interview Survey, confirmed those reports. Stuttering school children were significantly more likely to repeat a grade than control children. From six Italian schools, a report<sup>174</sup> studied 52 child stutterers and 374 controls, aged 8–17 years with a mean age of 11 years. Compared to the controls, there was evidence that the child stutterers had lower academic outcomes. There is evidence that children experience social isolation in school from fear of speaking in the classroom, and that they habitually avoid it.<sup>175,176,177,178</sup>

If I thought there was a teacher that would randomly pick kids to read or would go down the row and everybody gets a turn, I'd have my mother talk to them and once again explain my situation, so that I did not have to read in class because any time they started that my ears would get hot, I'd start getting nervous, I couldn't sit still, I just started to sweat, and the only thing I could think about was counting down the time until I had to read. (p. 77)

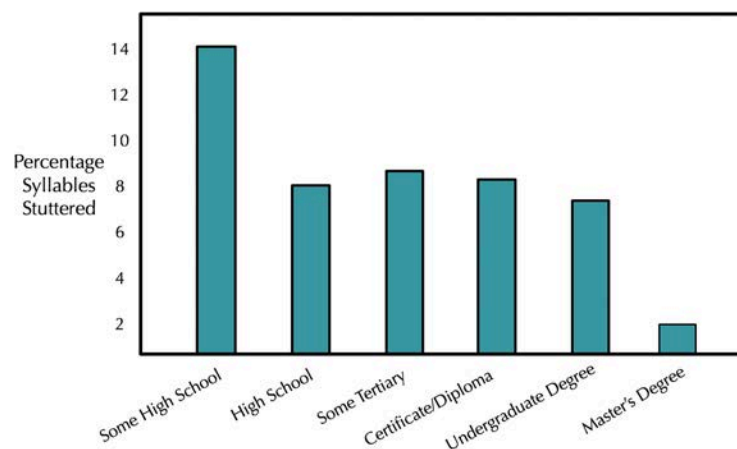
I used to go upstairs to the second floor bathroom and just keep flushing the toilet so that nobody could hear me getting sick, and then that kind of physical behavior lasted with me a long, long time because as a young adult and as an adult whenever I had a speaking situation coming up I would get physically ill. (p. 78)<sup>177</sup>

One student, they would read out a few stanzas. Then the next student would read out a few stanzas, and I could just tell immediately I was going to be number 22 there in the line. I just I couldn't think of anything else. And that moment of dread when my when it was my turn. And I hated that. I just hated that.<sup>32</sup>

The extensive clinical and theoretical aspects of this matter are explored in detail during Lectures Nine and Ten.

### *High school onwards*

The birth cohort study mentioned earlier<sup>166</sup> reported that stuttering had no effect on educational outcome. However, there is evidence that stuttering has a negative impact on education attainment. A study of 84 Japanese school students,<sup>179</sup> with a mean age of 14.6 years, reported that 26% of them failed to attend school for more than 30 days per year, which is much higher than the national average of below 1%. One report showed a negative linear relationship between stuttering severity and education attainment.<sup>180</sup> In other words, there is a tendency for those with more severe stuttering to attain less during education. Those data show that the stuttering of those who do not complete high school may be six times more severe than those who complete a postgraduate qualification. This is shown in the figure below.<sup>181</sup> The vertical axis gives a measure of stuttering severity (see Lecture Four). The horizontal axis shows various levels of education attainment, ranging from partial completion of high school to completing a master's degree at university. The effect in the figure—a significant negative correlation between stuttering severity and educational attainment—was replicated in a survey of 722 adults.<sup>105</sup>



One report<sup>182</sup> produced a finding that the websites of only 13% of 359 public universities provided information about alternative teaching and assessment methods for students who stutter, and only 51% of the disability liaison officers of those universities responded to an email enquiry about the topic. The authors pointed out that this could disempower potential university students who stutter because they cannot make informed choices about universities with pertinent disability services. Consequently, once at university, they might be unable to optimise their learning environments. A study of questionnaire responses of 20 adults in Jordan<sup>183</sup> showed that the following belief was ranked second on a list of 10: “your teachers/professors exclude you from participation in the classroom because of your stuttering.” A survey of 246 adult stutterers and 246 controls<sup>184</sup> in the United States reported that the former group perceived more negative perceptions from their college professors than controls. Additionally, the adult stutterers felt less comfortable approaching their professors.

## **Stereotypes and stigma**

### *A significant topic for stuttering*

When applied to people, the term “stereotype” refers to generalisations, which are usually simplified, and “stigma” refers to when such generalisations are negative. Many research publications have shown that those who stutter are affected by negative stereotypes about stuttering.<sup>185</sup> This appears to be true across the lifespan from childhood to adulthood.<sup>186,187,188</sup> The topic was considered of sufficient importance to warrant a conference about it.<sup>189</sup>

I guess the thing that happens most now is that if I stutter, people assume I'm anxious, shy, and may start to talk to me as if I'm a very small child. And that's

not appropriate. I can understand why they do it. They're trying to be helpful. They're trying to put you at ease, I suppose, but it's a bit condescending. So I don't like it.

### *Stereotypes about stuttering in fiction*

Stuttering is frequently depicted in movies and television with a negative stereotype.<sup>190</sup> A review of 29 works of fiction that contained a character who stuttered<sup>191</sup> indicated that “most often, characters who stuttered encountered mean-spirited teasing, name-calling, demeaning remarks or bullying from one or more of the characters” (p. 617). A detailed examination of stuttering portrayed in film<sup>192</sup> typically shows it to be used in a negative fashion: for example, as a comic device or as a sign of weakness.

### *Responses to stuttering*

A report<sup>193</sup> collected listener responses to speech read by participants who stuttered and controls. Without any identifying information about members of the former group, the listeners judged them to be less intelligent, less likeable, and more anxious than the controls. A study of 324 adults who stuttered from the United States,<sup>194</sup> involved a list of 15 stigmatising experiences. For most of these experiences, most participants reported having experienced them during their lifetimes. An interview with seven adult stutterers<sup>195</sup> raised the issue of those who stutter experiencing microaggressions, which are subtle manifestations of negativity about marginalised groups.

During adolescence, it seems that peers find those who stutter “nervous” and less attractive than others.<sup>196</sup> One of these findings<sup>197</sup> was that 736 adolescent and young adults stated that images of young people labelled as a person who stutters were less physically attractive than images without that label.

### *A cross-cultural issue*

There is compelling evidence that communities of various cultures hold negative stereotypes about those who stutter. A review of that evidence<sup>198</sup> stated,

the public view of stuttering is generally unfavorable and ... listeners often ascribe negative traits like anxious, shy, nervous, unassertive or introverted to people who stutter. (p. 54–55)

Since this review, publications have reported stuttering stereotypes in the United States,<sup>199,200</sup> Poland,<sup>201</sup> Japan,<sup>202</sup> and Chinese populations.<sup>203</sup> As noted earlier, this stereotyping extends to occupational suitability.<sup>161,162,163</sup> There is evidence that those who stutter may hold self-stigmatising thoughts in response to such community attitudes, and that those self-stigmatising thoughts may contribute to psycho-social harm.<sup>204</sup>

### *Stereotypes and speech-language pathologists*

Added to this literature is a recurring finding over several decades implicating speech-language pathologists in the perpetuation of negative stereotypes about stuttering. Clinicians in the United States,<sup>205,206</sup> the United Kingdom,<sup>207,208,209</sup> Korea<sup>210</sup> and Turkey<sup>211</sup> are reported to have negative and empirically unjustifiable attitudes to stuttering, although the research shows some signs of improvement of this problem over time. One report<sup>212</sup> raised an interesting suggestion that negative attitudes toward stuttering by speech-language pathologists are not necessarily revealed with standard, overt testing methods; there may be implicit attitudes that emerge with another means of testing.

### *POSHA-S*

A long-standing project has used a tool called the *Public Opinion Survey of Human Attributes-Stuttering* (POSHA-S) to explore worldwide attitudes to stuttering. A summary of a series of European reports<sup>213</sup> from 1,111 respondents found some variation, including less positive than average attitudes in Italy and more positive than average attitudes in Norway and Sweden. That international database was used to report that stuttering seems more stigmatised than obesity but less stigmatised than mental illness.<sup>214</sup> Subsequently, reports dealing with attitudes in Australia,<sup>215</sup> Turkey,<sup>216</sup> Portugal,<sup>217</sup> China and Japan,<sup>218</sup> Egypt,<sup>219</sup> and Malaysia<sup>220</sup> have been published. A report<sup>221</sup> showed that “Saudi people

generally hold sensitive attitudes toward PWS [*people who stutter*]” (p. 13), although knowledge about stuttering in that country seemed to be limited.

A report with the POSHA-S<sup>222</sup> showed that British, Arabic and Chinese students enrolled in a British university had stereotypical attitudes to those who stutter, and that their attitudes differed based on their home cultures. Another report<sup>223</sup> indicated that Chinese college students hold more negative attitudes to stuttering than United States college students. A report with the POSHA-S database<sup>224</sup> showed that protective services workers—police officers, fire-fighters, security guards—had more negative beliefs about those who stutter than other occupational groups. Using a different survey instrument, another group<sup>225</sup> reported that awareness and accurate knowledge about stuttering was limited in Japan. The POSHA-S data base was used to explore attitudes of parents with a child of any age who stuttered. Comparisons were made for the Middle East with Europe and North America.<sup>226</sup> All differences found were small, with only 4% of POSHA-S scores explained by variables investigated. However, parent attitudes in the Middle East were more negative than Europe and North America. Also, mothers in the Middle East had significantly better attitudes than fathers.

### *Dealing with stigma about stuttering*

Another report<sup>227</sup> presented information about the relative merits of different approaches to dealing with stuttering stigma, based on procedures used for dealing with stereotypes about mental illness. The first approach to dealing with stigma about stuttering is to provide public information about what it is like to experience the condition. The second approach is to provide public education based on the traditional “fact and fiction” approach. Finally, the protest approach draws attention to the injustice and inappropriateness of stigmatising a condition. The report found that all three approaches had value for reducing stereotypes about stuttering.

### *Stereotypes and teachers*

Some reports suggest that United States teachers of children younger than 12 years,<sup>228,229,230</sup> special educators in schools,<sup>231</sup> and school administrators<sup>232</sup> may well hold negative and unjustifiable stereotypes about stuttering. More recent reports from the United States<sup>233</sup> and India<sup>234</sup> were more favourable, although another report showed that United States teachers did not have more accurate beliefs about stuttering than the public.<sup>235</sup> A study of New Zealand teachers<sup>236</sup> found they did not have negative attitudes about stuttering, but they did have knowledge gaps about its causality (causality is discussed in Lecture Three). An Australian study<sup>237</sup> involved 51 student teachers of children 12 years and younger. Of those teachers, 75% agreed that the cause of stuttering was “underlying psychological problems” (p. 714), and 71% agreed that it was “a combination of physical and psychological problems” (p. 715). Seventy-two percent agreed that children that age were likely to be shy, and 75% agreed that they are likely to be anxious. A study of 74 early childhood educators in Greece<sup>238</sup> reported that they had overall positive attitudes toward 2–5-year-olds who stuttered. The report also indicated a relationship between knowledge about stuttering and positive attitudes to it.

A report<sup>239</sup> of 262 Kuwaiti teachers and 209 trainee teachers found, among other things, that 81% of the teachers and trainee teachers believed stuttering to be caused by emotional problems. Additionally, 76% believed it to be precipitated by “a very frightening event,” and 15% believed that “a virus or disease” (p. 60)<sup>239</sup> was responsible. Additionally, 20% of them indicated that “people who stutter should try to hide their stuttering,” 72% believed those who stutter to be “nervous or excitable,” 82% believed they were “shy or fearful,” and 35% indicated that those who stutter “have themselves to blame for their stuttering” (p. 61).<sup>239</sup> A report around the same time<sup>240</sup> indicated that a third of Kuwaiti teachers reported negative attitudes to stuttering, with a more positive attitude for United States teachers. A follow-up study<sup>241</sup> showed that such stereotypes could be corrected during teacher training with a 17-minute educational video about stuttering. A report of interviews with 10 Belgian teachers of adolescent students<sup>242</sup> showed that they felt stuttering could become a problem if attention is given to it, and that they tried to minimise any reaction to stuttering and rarely talked about it in class. A report of 106 Jordanian special education teachers<sup>243</sup> indicated that they had limited knowledge about stuttering. A report of 55 Australian primary school teachers<sup>244</sup> indicated that three-quarters of them believed that stuttering has psychological causality and that students who stutter are

likely to be anxious or shy.

### *Stereotypes and universities*

There is some evidence that stereotypes about stuttering extend to university environments. University students have been shown to have negative attitudes to stuttering.<sup>245,246</sup> One report<sup>247</sup> showed that university professors and students scored students who stutter as having more negative personality traits than other students. However, another report<sup>248</sup> of student perception failed to find overriding stereotypes, and another found “neutral to positive perception” (p. 206)<sup>249</sup> of students who stuttered. Speech-language pathology students have been shown to have more positive attitudes toward stuttering than others, with some evidence that there may be differences across countries.<sup>250</sup> A report of students from an Australian university<sup>251</sup> indicated a positive attitude toward stuttering and suggested a connection between that result and curriculum content. An experimental report of professor evaluations of oral student presentations found a that, in some situations, professors may compensate for students who stutter.<sup>252</sup> This occurred in cases where a student stuttered and had poor communication skills.

### *Self-disclosure and stereotypes*

Some researchers<sup>253</sup> have made the important point that, in addition to being stigmatised, stuttering is not always readily apparent because it can be so variable from situation to situation (as discussed earlier in this lecture). Further, it can be actively concealed, such as by means of “covert stuttering,” which is described shortly. With a survey of 505 adult stutterers, the researchers presented data showing that the extent to which stuttering was concealed adversely impacted quality of life.

The opposite of concealment is to self-disclose. In the case of stuttering, self-disclosure is when, at the start of a social interaction or speaking to listeners for the first time, speakers declare that they stutter. There is evidence that self-disclosure can positively influence listener reaction to stuttering,<sup>254</sup> with only one report of no effect.<sup>255</sup> University professors have been shown to view students more positively when they self-disclose.<sup>256</sup> Self-disclosure by a 12-year old and his teacher was shown to be beneficial.<sup>257</sup> A study reported that favourable perceptions about a 12-year-old were associated with a written disclosure statement by his mother, rather than by his teacher or by himself.<sup>258</sup> There is some evidence to associate self-disclosure with speaker perceptions of speech spontaneity.<sup>145</sup>

Much of the literature about self-disclosure suggests that the procedure might be used to deal with anxiety about speaking. For example,

It helps reduce anxiety and nervousness because it addresses the elephant in the room. So, it helps in that way, which makes me less nervous for the conversation to come, especially if it's an important one like an interview. (p. 2051)<sup>259</sup>

In which case, from the perspective of clinical psychology, there is a potential clinical reservation about its use (a potential safety behaviour), which is discussed during Lecture Ten.

### *Self-stigma and stuttering*

There is a distinction between public stigma and self-stigma.<sup>260</sup> According to those authors, the former term refers to endorsement of stereotypes by a general population, but self-stigma refers to when people internalise such stereotypes, applying them to themselves. With stuttering, an early study<sup>261</sup> prompted a suggestion that “the expectation creates the reality, or, in other words, the stigma shapes the stutterer” (p. 487). More recent data<sup>262</sup> support this contention. A survey of 344 adult stutterers used regression modelling to show that various measures of self-stigma significantly predicted measures of global mental health and how much stuttering interfered with communication. A report with 101 participants 10–18 years<sup>263</sup> indicated that self-stigma correlated with the impact of stuttering and showed that self-stigma may be present in 10-year-olds.

## Anticipation of stuttering

### *A common effect*

It has been known since the 1930s that those who stutter anticipate its occurrence with some reliability.<sup>264</sup> This knowledge has been bolstered by reports with adults<sup>265</sup> and children<sup>266,267,268</sup> during reading tasks that have established eye gaze patterns consistent with anticipation of difficulty with certain words. All this knowledge has figured in many of the influential causal explanations about stuttering in the history of thought about it (see Lecture Three): primary and secondary stuttering theory, the Diagnosogenic Theory, approach-avoidance theory, and the Anticipatory Struggle Hypothesis.

### *The experience of anticipating stuttering*

Those who stutter commonly report anticipation of stuttering. A study of 30 adults<sup>122</sup> reported their experiences of anticipating stuttering, and around half reported “they experience anxiety or uncertainty when they anticipate stuttering” (p. 44). All reported using at least one proactive response to the feeling of anticipating stuttering. For example, “an attempt to hide or escape from an impending moment of stuttering” (p. 42) was reported by 87% of participants. Circumlocution, and including something in a conversation that was not originally intended, was the most common avoidance response.<sup>†</sup> Consistent with that report, word substitution was independently reported by 82% of another cohort of stuttering participants.<sup>269</sup> Avoiding situations was also a common proactive response to anticipating stuttering.

Self-management strategies, either learned in a clinic or self-generated, were reported by 87% of participants.<sup>269</sup> Those included variants of the speech restructuring technique to be discussed later this lecture, relaxation procedures, and reducing speech rate. Forty per cent of participants reported consciously deciding to not alter speech in any way in response to a feeling of anticipation. The participants indicated that the experience of anticipating stuttering can be helpful to them but also harmful, with 43% reporting that it can be both. However, 37% reported that it is of no help at all and a minority of 13% reported that it is always helpful.

Anticipation of stuttering is connected to what is commonly referred to as *covert stuttering*,<sup>270</sup> and sometimes *interiorized stuttering*.<sup>271</sup> These terms refer to attempted stuttering concealment using techniques such as word avoidance, circumlocution, and situation avoidance. It seems that it is common for those who stutter to pass through a stage where they initially practice covert stuttering but eventually abandon those efforts.<sup>272,273</sup>

Currently there is some doubt about the impact of covert stuttering on the person overall. A report of 21 adult stutterers<sup>274</sup> categorised them as having “predominantly overt stuttering features” and “predominantly covert stuttering features” (p. 8). No differences were reported between the groups in terms of how stuttering affected the participants. However, another survey of 502 adult stutterers<sup>275</sup> categorised them as those who predominantly “have a goal of not stuttering when speaking” and those who predominantly “have the goal of stuttering openly and trying not to hide stuttering” (p. 4339). Those in the latter group were reported to less often experience negative emotions about their stuttering.

Research has begun to develop an instrument to measure anticipation of stuttering events: the Premonitory Awareness in Stuttering Scale.<sup>276</sup> This 12-item scale was adapted from a similar scale used for tics, and showed that adults who stutter report anticipation of “speech disruptions” more often than control speakers.

There is some evidence of a physiological basis to anticipation of stuttering.<sup>277</sup> Forty adults who did not stutter listened to audio recordings of utterances by adults who stuttered. Thirty-nine of the utterances that had a stuttering moment partway through or at the end were edited to stop before the

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<sup>†</sup> Clients commonly report that this can be a functional issue, such as not ordering a particular menu item in a restaurant to avoid stuttering while giving the order to the waiter.

stuttering moment. Forty-nine matched utterances did not contain a stuttering moment. There was statistical evidence to suggest that the listeners may have been able to anticipate which utterances would contain stuttering and which would not. The experimenters concluded that this must have been because of acoustic information.

## Social anxiety

### *Situation avoidance*

The effects of stuttering in the ways just discussed—speech output, occupational and educational attainment, social stereotypes, and situation avoidance—are probably connected to a common effect of stuttering. That effect is social anxiety and is considered in detail during Lectures Ten and Eleven. As will be discussed then, a common effect of social anxiety is to avoid speaking situations.

An early report<sup>278</sup> documented situations that are commonly avoided by those who stutter, using 50 stuttering participants prior to treatment and 100 controls. They indicated their avoidance of 40 standard speaking situations. The following table\* presents the top 15 situations that were avoided by the groups, with the most avoided situations at the top of the list. The ranking is ordered according to the stuttering participants.

<i>AVOIDED SITUATION</i>	<i>STUTTERING</i>	<i>CONTROLS</i>
<i>Asking a question in class</i>	1	2
<i>Speech to unfamiliar audience</i>	2	1
<i>Telephoning to make enquiries</i>	2	19
<i>Short class recitation</i>	4	8
<i>Reading aloud to friends</i>	5	14
<i>Introducing one person to another</i>	6	18
<i>Introducing oneself</i>	7	7
<i>Telephoning for a meeting or appointment</i>	8	24
<i>Parlour games requiring speech</i>	9	10
<i>Telephoning for a taxi</i>	9	26
<i>Giving your name over the phone</i>	11	21
<i>Asking for a job</i>	12	6
<i>Participating in committee meetings</i>	13	12
<i>Telling a joke to a stranger in a crowd</i>	14	5
<i>Giving someone a message</i>	15	27

Most obviously from the table, the telephone was a recurring avoided situation reported by those who stuttered compared to controls. It also seems that those who stutter avoided group speaking situations more often than controls. The report also showed that those who stutter were the most comfortable with people they knew, such as friends and family, and did not commonly avoid those situations.

A more recent publication<sup>279</sup> used another situation checklist<sup>280</sup> and compiled data from 88 adult participants seeking treatment for stuttering and 209 controls. The checklist includes items dealing with emotional responses such as anxiety and worry, and items dealing with the “likelihood of speech breakdown.” For emotional responses, the three items that best distinguished the stuttering and control participants were “talking on the telephone,” “asking a teacher or supervisor a question,” and “being

\* Adapted and reproduced with permission: Trotter, W., & Bergmann, M. (1957). Stutterers' and nonstutterers' reactions to speech situations. *Journal of Speech and Hearing Disorders*, 22, 40–45. © 1957 American Speech-Language-Hearing Association.

rushed." For likelihood of speech breakdown, the three items that best distinguished the two groups were "talking on the telephone," "talking with teachers or supervisors," and "saying a sound or word that previously has been troublesome" (p. 1137).<sup>279</sup>

### *The telephone*

The problematic nature of talking on the telephone for those who stutter is further shown by that speaking situation being at the top of their hierarchies of feared and avoided situations.

Whenever possible I ask my dad to use the phone for me because using the phone really, really scares me. But recently I've been trying to answer calls myself and I have been becoming braver, which is really good.

In my earlier working days, I would prefer to walk up three flights of stairs rather than to make a phone call.

If I had a bad block, when someone would phone, I would pick it up and 40 seconds later I could make no noise. The people would assume that there was a problem with the phone and they would hang up. Very frustrating and demoralising.

The person on the other end of the line doesn't know that I'm stuttering or I'm having a block. So to them it may come across as I'm not paying attention to them or I'm not answering their question.<sup>32</sup>

One report was a survey of 223 British participants.<sup>281</sup> Those who rated their stuttering to be severe reported making fewer telephone calls per week than those with milder self-ratings of severity. Thirteen per cent of participants reported always using an alternative to the telephone and 55% reported sometimes doing so. In this report more than a third of those with self-reported severe stuttering said they always used alternatives to the telephone, and more than half reported sometimes having others make calls for them. Sixty per cent agreed with the statement that "it is more difficult to speak to someone on the 'phone than 'face-to-face'" (pp. 308–309).<sup>281</sup> Recurring reasons given for this were that nonverbal communication is not possible by telephone, reactions to stuttering are unknown on the telephone, a lack of understanding of stuttering by the conversation partner, and time pressure. Generally, making calls was reported to be more troublesome than answering them. Compared to participants older than 50 years, twice as many participants younger than 30 years reported always using alternatives to telephoning.

Another study was an interview report of 130 stuttering participants.<sup>282</sup> They were asked "of all your feared talking situations, where would you rank calling on the telephone?" (p. 235). They were also asked about answering the telephone. Overall, 72% ranked making calls among their top three feared situations, and 54% made that rating for answering the telephone. As with the previous report<sup>281</sup> those effects were much more pronounced for severe cases. Participants were given a list of telephone calling options to rate on a fear scale, and the following were the most highly rated: someone from a different culture, the opposite gender, directory assistance, telephone operator, store enquiry, and an older person.

An interesting way to gain insight into these issues has been reported<sup>283</sup> by faking stuttering—often referred to as pseudostuttering—while telephoning a stranger, such as a travel agent or a department store staff member. Twenty-nine graduate speech-language pathology students who did this found the experience rather sobering, with evidence that it may have promoted negative self-perceptions.

### **Personality**

A well-known reference text<sup>284</sup> concluded that "there would seem to be some justification for the inference that stutterers on the average are not quite as well adjusted as are typically fluent speakers" (p. 308). Publications since that review confirm an impression that those who stutter may have unusual personalities compared to those who do not stutter. A report with the Minnesota Multiphasic Personality Inventory<sup>285</sup> showed significant differences between the two groups. Another report<sup>286</sup> compared 93 adults seeking treatment for stuttering with matched controls using a test called the NEO



Five Factor Inventory, which assesses five personality domains: Extraversion, Neuroticism, Openness, Agreeableness, and Conscientiousness. Results showed that the stuttering participants were all within the typical range for the five domains but had higher Neuroticism and lower Agreeableness and Conscientiousness scores than controls.

Another report<sup>287</sup> using the same assessment and groups of around the same size of 87, replicated that initial report about Neuroticism but found the opposite for Conscientiousness and Agreeableness, reporting higher scores than controls. Using the NEO Five Factor Inventory again, another report<sup>288</sup> was consistent with the Neuroticism finding by reporting that it correlated with high impact of stuttering among those who stutter.<sup>†</sup> However, using the same measure with a culturally different population, another study<sup>289</sup> found only higher Agreeableness scores for the stuttering group, but no other differences. Two reports have also linked stuttering to perfectionism.<sup>290,291</sup>

In short, there are inconsistent and slight differences found across studies. Possibly, this is because some personality disorders can be explained simply as the effects of stuttering, and some cannot.<sup>292</sup> These generally inconclusive findings about stuttering and personality are consistent with the inconclusive results of a study<sup>293</sup> dealing with a related construct: temperament. The researchers concluded that, from among 13 aspects of temperament, there is “a nontrivial tendency for AWS [adults who stutter] to experience decreased positive affect compared to AWNS [adults who do not stutter]” (p. 2691). The topic of temperament and stuttering during the early years of life is covered in detail in Lecture Eleven.

### Qualitative research about the lived experience of stuttering

There is a body of literature involving interviews with those who stutter to establish information about their experiences with it. A synthesis of such reports since 2000<sup>294</sup> involved 17 studies that met methodological criteria. The authors reported that five themes figured prominently in that literature:

- (1) Avoidance is used to manage stuttering ...
- (2) Stuttering unfavourably impacts employment experiences ...
- (3) Stuttering shapes self-identity ...
- (4) Stuttering leads to negative reactions ... both actual and perceived ...
- (5) Stuttering impacts relationships adversely. (p. 2237–2239)<sup>294</sup>

## GUIDELINES FOR INTERACTING WITH THOSE WHO STUTTER

### An important topic

According to one publication, there is no shortage of recommendations to the public about how to interact with those who stutter.<sup>295</sup> Yet, as noted in that publication, little of that advice has been generated by those who stutter. Consequently, the authors elicited the views of 148 adults, most of whom had received treatment or support from a self-help group for stuttering. Two thirds were men. From a list of 24 items, the following three actions were rated most highly supportive, in this rank order:<sup>295</sup>

- (1) “maintain eye contact”
- (2) “wait to let a PWS [person who stutters] say what he/she wants to say”
- (3) “assuming the listener also stutters, to ask the PWS how they can help the listener with his/her own stuttering problem” (p. 5)



<sup>†</sup> Neuroticism is a tendency to experience high levels of negative emotion such as anxiety, anger and sadness. Impact of stuttering was measured with the OASES, which is discussed in Lecture Four.

The following three actions were rated as least supportive:

- (1) “‘faking’ stuttering during conversation”
- (2) “telling the PWS [*person who stutters*] how he/she should feel about the problem”
- (3) “trying to ‘help’ the PWS by finishing stuttered words” (p. 5)

From written responses to a question about desirable and undesirable responses during communication, the following nine actions were listed as supportive by at least 10 respondents, in this rank order:<sup>295</sup>

- (1) “maintaining eye contact”
- (2) “being patient, understanding, sensitive, friendly or non-judgemental”
- (3) “listening”
- (4) “asking about stuttering”
- (5) “allowing the PWS [*person who stutters*] to finish his or her words or sentences”
- (6) “showing empathy, interest, compassion, or respect”
- (7) “treating the PWS normally”
- (8) “engaging him or her in conversation”
- (9) “helping with the stuttering” (p. 8)

Ten or more respondents considered these two actions not supportive:<sup>295</sup>

- (1) “finishing one’s words or sentences”
- (2) “ridiculing one’s stuttering (e.g., making fun or mocking)” (p. 8)

### Two caveats about eye contact

The authors presented two caveats about the recurring finding (shown in the list above) that respondents indicated eye contact to be the most supportive action by conversational partners. First, it may cause discomfort during conversation with someone who stutters by creating a feeling of “staring.” Second, eye contact is not desirable in some cultures.

## CONDITIONS THAT REDUCE OR ELIMINATE STUTTERING

### The fluency inducing conditions

#### *Changing customary speech*

A fascinating feature of stuttering is that speakers can change how they speak in certain ways, and this can reduce stuttering or even get rid of it completely while they are using those speech changes.<sup>296</sup> Sometimes the changes can be subtle. For example, it is common to hear of actors who stutter but do not do so when they are on stage. Presumably, part of the explanation for this is the change to customary speech while on stage: louder, slower, perhaps with a different accent, and so on.<sup>†</sup>

#### *The Modified Vocalisation Hypothesis*

The term “fluency inducing conditions” is attributed to Wingate, who proposed a Modified Vocalisation Hypothesis to explain why some speaking conditions reduce stuttering.<sup>297</sup> Wingate proposed that “speaking under all of these conditions emphasizes vocalization and continuity of vocalization” (p. 682).<sup>297</sup> There is much research about these conditions, which is outlined in a reference text (Chapter 11).<sup>284</sup> Some of the common fluency inducing conditions are described below.

#### *Singing*

Arguably the best known feature of stuttering is that it goes away during singing. There are some who question whether this always happens (p. 425),<sup>298</sup> but it is generally accepted as a feature of stuttering.

<sup>†</sup> Another part of the explanation is that actors on stage know what they are about to say; there is nothing spontaneous about their speech.

Singing has never been directly linked to the development of a treatment method. However, an acoustic analysis of adolescents who stutter and controls during singing<sup>299</sup> showed changes consistent with a popular treatment method to be discussed shortly: *speech restructuring*. In short, singing stabilises and simplifies speech motor activity. Acoustic changes associated with singing have been summarised:<sup>300</sup>

During singing, relative to speaking, the temporal structure and the coordination of laryngeal and oral movements are altered (i.e., the proportion of short phonation intervals is reduced, vowel durations are lengthened ... articulation rate is slowed ... and articulatory voicing is stabilized (p. 11)

And, as with acting, the social context of singing is different from everyday speech. The words of a song are not spontaneous but are known in advance.

### *Rhythmic speech*

Speaking in time to a rhythm has a similar universal effect on stuttering. This has been the source of many therapy techniques during past decades, and even past centuries according to common belief. Writings by Plutarch in 75 BC convey that, Demosthenes, a famous Greek orator of the third century BC, stuttered. He consulted the Greek actor Satyrus, who prescribed (among other treatments) that Demosthenes should run or walk uphill while speaking.<sup>301</sup> This is commonly interpreted as the first therapeutic use of rhythm to reduce or eliminate stuttering.

Since then, there have been many clinical applications of rhythmic speech. Many have not been particularly successful, or they were ethically dubious. An example of the latter are the now infamous “stuttering schools” that proliferated in the United States during the first part of the 20<sup>th</sup> Century.<sup>302</sup> Miniature in-the-ear metronome devices also emerged during the 1970s<sup>303</sup> but never attained any empirically verified success.

There has been a great deal of research about the rhythm effect, dating from the first half of the last century.<sup>304</sup> This body of research is too detailed to review in the context of these lectures. However, a recent, sophisticated functional magnetic resonance imaging report<sup>305</sup> provided the first neurophysiological details about how rhythmic speech might control stuttering. There has been a sophisticated acoustic analysis of how rhythmic speech differentially affects the timing of syllables for adolescents and school-age children who stutter and controls.<sup>306</sup> There is evidence that adults who stutter show poorer discrimination of complex rhythms than adults who do not stutter.<sup>307</sup>

### *Reduced speech rate*

Virtually everyone who stutters will report being told at some time to “slow down.” This advice is presumably based on an assumption by casual observers that the problem of stuttering arises from attempting to speak too quickly. Reduced speech rate is a component of many modern treatments.

Of itself, however, the speech rate reduction needed to attain clinically useful stuttering reduction may not be functionally useful. This contention is supported by a report<sup>308</sup> that a 30% reduction of reading rate did not significantly reduce stuttering. For the severest of the participants, the 30% speech rate reduction reduced stuttering severity by 35%, leaving the participants with considerable stuttering.

### *Chorus reading and shadowing*

When someone who stutters reads in chorus—that is, at the same time—with someone who does not stutter, stuttering disappears during the reading. Even more curious, if someone who stutters repeats what another person has just said during a spontaneous monologue, but a few words later, the same thing occurs. The former condition is called *chorus reading* and the latter condition is called *shadowing*. There is some evidence that the Modified Vocalisation hypothesis could explain the chorus reading effect.<sup>309</sup> There is also evidence that chorus reading might be explained by an effect where the speech pattern of the reader who stutters is influenced by the reader who does not stutter.<sup>310</sup> Neither of these speaking conditions has influenced modern treatment practices.

## Verbal response contingent stimulation

### *Response contingent stimulation research with stuttering*

During the early 1950s, and ending some decades later, there was a series of laboratory experiments involving aversive stimulation with electric shock or loud noise. When shock or loud noise occurred after stuttering moments, stuttering decreased, and in some cases stopped altogether, only to return when the shock or noise ceased. There are at least 50 publications to that effect, dating from the early 1960s.<sup>311</sup> The shock or noise is called *response contingent stimulation of stuttering*. The results of those experiments show that stuttering has operant features.

### *Stuttering has operant features*

It is important to state that stuttering has operant features, not that stuttering is an operant. If a behaviour is freely emitted and readily controllable, and changes with response contingent stimulation, then the behaviour is referred to as an operant. However, stuttering is not a freely emitted problem behaviour. As will be discussed during Lecture Three, stuttering is atypical neural processing of speech, which is beyond the control of those affected. A treatment for stuttering that incorporates response contingent stimulation can be referred to as a treatment with operant methods, or an operant treatment.

### *Verbal response contingent stimulation of stuttering*

Laboratory research of shock and loud noise with stuttering stopped during the mid 1970s. However, it did lead to the discovery that response contingent stimulation of stuttering could be verbal, and could functionally control stuttering.<sup>312,313,314</sup> Research showing that this was an option for children<sup>315,316,317</sup> established clinical possibilities that have been fruitful, particularly for treatment of early childhood stuttering, as will be discussed during Lectures Six and Seven. In short, a treatment based on parent verbal response contingent stimulation—the *Lidcombe Program*—has been shown with many clinical trials to be efficacious.

There is some evidence, albeit patchy,<sup>318,319</sup> that the Modified Vocalisation Hypothesis might explain the verbal response contingent stimulation effect. There is also evidence, again not particularly compelling, that an explanation might be that the verbal response contingent stimulation reduces stuttering by inducing simplifications to spoken language.<sup>320,321</sup>

## Auditory feedback

### *Delayed auditory feedback, altered auditory feedback, and speech restructuring*

*Delayed auditory feedback and altered auditory feedback* refer to when airborne speech feedback is altered with an electronic device by means of unilateral or bilateral headphones or an in-the-ear device like a hearing aid. The first such effect to be discovered was *delayed auditory feedback*,<sup>322,323</sup> often referred to as DAF. After a famous report of it being used to reduce stuttering,<sup>324</sup> this discovery prompted much research that continues to the present and has profoundly influenced treatment practices. Generally, delayed auditory feedback creates a slow and unusual drawling speech pattern that reduces or eliminates stuttering. This is the basis for the most popular of modern treatments for reducing the stuttering of adolescents and adults who stutter. It is rare for delayed auditory feedback devices to be used clinically these days, and clinicians simply teach those who stutter how to use a novel speech pattern to reduce or eliminate stuttering. These treatments, discussed during Lecture Eight, are referred to generically as *speech restructuring*.<sup>325</sup> In short, speech restructuring treatments have been shown with many clinical trials to be efficacious.

For nonstuttering speakers, delayed auditory feedback can induce disfluencies that once were thought to resemble stuttering,<sup>326</sup> and this prompted many propositions that stuttering was caused by a problem with speech feedback. However, it is now accepted that these disfluencies are not stuttering. Potentially, delayed auditory feedback devices are problematic, because there have been reports of them<sup>298</sup> inducing transient speech problems (pp. 372–373).

Altered auditory feedback devices are a modern development of delayed auditory feedback. In

addition to delaying speech feedback, these devices alter pitch upwards or downwards. Such devices are commercially available, but their clinical value appears to be questionable at present, as discussed during Lecture Eight.

### *Masking*

Stuttering is significantly reduced or eliminated when the speaker's voice is not fed back because of noise—commonly white noise—presented through earphones. To return to Demosthenes, there are some sources that suggest Satyrus prescribed that Demosthenes also practise speaking on the seashore above the noise of a roaring ocean. It is tempting to speculate that Satyrus thus discovered and found a clinical application for the masking effect in, addition to the rhythm effect.

### **Speaking alone**

Intriguingly, historical textbooks in the field have claimed anecdotally that a defining feature of stuttering is that it does not occur when speaking alone without a listener.<sup>327,328,329,330</sup> Empirical reports confirm that there will be more stuttering when speaking to people than when speaking alone.<sup>331,332,333</sup> However, only one study has observed participants speaking when they are deceived into believing they are speaking alone.<sup>334</sup> Under those conditions, almost no stuttering occurred. There is much experimentation needed about this effect because of its potential clinical importance. As discussed during Lecture Ten, social anxiety is strongly associated with stuttering. Further laboratory experimentation using control groups is needed to determine whether the speaking alone effect occurs because social anxiety is removed altogether, or whether it occurs for other reasons. As the authors of the latter paper note,<sup>334</sup> linguistic changes to customary speech while talking alone, or changes to speech motor function, could account for the effect. There is some evidence consistent with the latter prospect.<sup>335</sup> That study linked external focus on an audience with reduced variability of lip aperture during speech.

### **SUMMARY**

Stuttering can be associated with potentially confusing terminology that is best to avoid. It is a clinically useful idea that stuttering involves moments that interrupt speech. There is no all-purpose definition of stuttering, but three common definitions can be used in different clinical contexts. The observable behaviours of stuttering are many and complicated, so it is clinically important to have ways to describe them clearly. The distribution of stuttering moments during spoken language is generally influenced by initial word consonants, and those who stutter commonly find that certain sounds are often stuttered. There is evidence that stuttering affects quality of life as much as chronic health conditions such as diabetes mellitus, cardiovascular disease, cancer, HIV, and chronic kidney disease. Presumably, that impact is caused because, unlike those health issues, stuttering develops in the first years of life. Stuttering has direct monetary costs such as treatment expenses and lost work hours, and indirect costs to society involving education, occupation, and restricted social functioning. Those who stutter are marginalised in society by negative stereotypes and stigma. Stuttering commonly causes social anxiety, which is connected to situation avoidance. There are well established guidelines for how to interact with those who stutter. There are many conditions that reduce or eliminate stuttering, and many of those are used in successful treatment methods.

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## LECTURE TWO: MORE BASIC INFORMATION

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### EARLY AND PERSISTENT STUTTERING: A FUNDAMENTAL CLINICAL DISTINCTION<sup>†</sup>

#### Early stuttering

##### *Overview of early childhood stuttering*

For the purposes of these lectures, the term *early childhood stuttering* refers to a stage of its development during which there is a chance that recovery will occur, either because of natural recovery, treatment, or a combination of both. The period of early childhood stuttering begins at onset and extends for some years after that.

##### *Tractability of early childhood stuttering*

Early stuttering appears to be extremely tractable; in other words, it is responsive to treatment. Many children recover naturally without any formal treatment, as discussed later during this lecture. It is possible—even likely—that early treatment facilitates natural recovery.<sup>1</sup> As will be discussed during Lecture Nine, it seems that stuttering starts losing clinical tractability at some time during the period when children are 6–11 years old. A review of treatment reports available at the end of the last century<sup>2</sup> concluded this about the need for early treatment: “treatment after more than 15 months have elapsed does not appear to have been as effective ... as treatment initiated sooner” (p. 223).

##### *Parent contact with children*

Formal education in most English speaking countries begins between the ages of 4 to 6 years. Parents generally have most contact with their children prior to this age. It is true, though, that many children with early stuttering will attend pre-school, kindergartens, or day care centres, for all or part of the week. This is clinically relevant because parent contact with children during each day is a clinically essential feature of early stuttering treatment. Parents do the treatments outlined during Lecture Six when they are with their children.

##### *Early treatment is the best option*

Lecture One outlined how stuttering may be associated with negative feelings about loss of control and expectation of stuttering. As discussed during that lecture, educational and occupational limitations are often encountered by those who stutter, and those limitations are likely to be associated with social anxiety. As will be discussed during Lecture Eleven, there is good reason to believe that negative peer conditioning during the years of early childhood stuttering is implicated in the origins of social anxiety for those who stutter. Considering this, and considering that stuttering is at its most tractable during that period of life, early treatment is clearly the best option.

#### Persistent stuttering

##### *Overview of persistent stuttering*

For the purposes of these lectures, the term *persistent stuttering* refers to a stage during which there is no reasonable chance that recovery will occur, either because of natural recovery, treatment, or a combination of both.

Persistent stuttering lasts a lifetime, and, after a period of early childhood stuttering, may worsen throughout life. This is apparent from comparing data sets that measure stuttering severity during the years of early childhood stuttering and during adulthood.<sup>3</sup> The majority of studies suggest that stuttering, and the various problems associated with it, do not abate with advancing age.<sup>4,5,6,7</sup> However, there have been reports that stuttering decreases in prevalence and severity with older

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<sup>†</sup> Thanks to Robyn Lowe, Sue O'Brian, Ann Packman, and Ross Menzies for assistance with this material.

age.<sup>8,9</sup> A cross-sectional survey<sup>10</sup> of 852 adults, with a mean age of 49 years, and 135 children, with a mean age of 11 years, collected self-reported severity ratings using a 9-point scale. Results showed a pattern of increased scores up to 18 years of age, followed by a systematic decrease of around two scale values during subsequent decades of life (Figure 2, page 6). A study of 3,100 stuttering moments from 147 adolescents and adults<sup>11</sup> reported that, during that span of life, stuttering became more behaviourally complex. With advancing age, repeated movements, fixed postures, and superfluous behaviours tended to appear.

#### *Treatment for persistent stuttering*

Treatment for persistent stuttering is designed to help clients control stuttering or deal with its effects, or both. Those affected by persistent stuttering may or may not seek such treatment or any form of support to deal with its effects.

## IDENTIFYING STUTTERING

### Clinical identification of stuttering

#### *Persistent stuttering*

Generally, clinicians don't need to diagnose stuttering in a clinic and tell people that they have it. Those who have stuttered for much of their lives will be fully aware of it. So, those with persistent stuttering who present to clinics seeking help will nearly always be correct that they stutter. The only clinical task is to confirm the presence of stuttering rather than some other condition, as discussed shortly.

#### *Early stuttering*

During the 1980s and 1990s many protocols were developed for distinguishing between stuttering and typical disfluency during the early years of life.<sup>12</sup> This topic was considered so important that a prominent clinical journal published two reviews in this area during the early 1990s.<sup>13,14</sup>

However, during this century there have been no further empirical developments or reviews published about differential diagnosis, which might reflect that such protocols are currently thought to be clinically unnecessary.<sup>15</sup> It seems that a case history and observation of a child's speech is sufficient to confirm early stuttering when parents bring children to a clinic. Some authorities in the field, after an earlier attempt to develop a differential diagnostic protocol, have endorsed such an opinion:

In our experience, the identification of early stuttering in clinical settings is seldom difficult. We wonder why several authors ... have expressed a different opinion, emphasizing the great overlap and possible confusion between early stuttering and normal disfluency, and cautioning clinicians of the difficult task. (p. 214–315)<sup>19</sup>

A report published shortly after this statement confirmed it. Child stutterers and a control group were studied speaking with parents and clinicians at home and at the clinic, with the conclusion that

... a clinician could, with some degree of confidence, predict whether a diagnosis of stuturer or nonstuturer based on a typical clinician–child conversation in a clinical setting would hold true in other environments. (p. 208)<sup>16</sup>

Consistent findings attesting to the ease of stuttering identification during its early stages emerged from a study of nine 3–5 year old Icelandic speaking children.<sup>17</sup> The researchers divided 7-minute speech samples from each of them into 5-second intervals and presented them to English and Icelandic clinicians. Neither group had any difficulty identifying which of the 5-second speech samples contained stuttering.

#### *Definition of stuttering and clinical identification of stuttering*

Lecture One stated that all three stuttering definitions—objective, internal, and perceptual—have

limitations, but they all have strengths that make them useful in different professional contexts. One of those contexts is when clinicians need to confirm early stuttering when parents bring a child to a clinic. In that situation, a combination of all three definitions can be applied to the task. The clinician can observe the repeated movements, fixed postures, and superfluous behaviours outlined during Lecture One. The clinician can use an internal definition when asking the parent if there are any signs that the child might be having negative experiences that are typically associated with stuttering. As considered during Lecture Eleven, such signs of negative experiences often do occur at stuttering onset. And finally, a perceptual definition can be used to determine whether the child's speech is consistent with other childhood stuttering that the clinician has observed.

#### *During Early identification errors*

There have been no studies designed specifically to determine how often there is a stuttering identification error when very young children are brought to clinics. However, one discussion of early stuttering identification noted of one specialist clinic that:

1,140 assessments for stuttering were conducted during the period 1994 to 2000, and a file audit showed that only 10 preschool children referred during that period (0.9%) were not identified to be stuttering. (p. 25)<sup>15</sup>

So, if that information is correct, for every 100 children brought to experienced clinicians for a consultation about stuttering, there would be uncertainty about one of them.

There are sources of anecdotal support from experienced clinicians and researchers about the accuracy of parent stuttering identification.<sup>18,19</sup> For example, a prominent authority stated, "indeed, I can recall only a handful of parental misdiagnoses of early childhood stuttering in more than 35 years of clinical practices in its identification and treatment" (p. 6).<sup>18</sup> Another authority stated that "typically, parents of young children who stutter correctly diagnose the problem, making the professional evaluation a task of describing and quantifying stuttering rather than differentiating it from other disorders" (p. 313).<sup>19</sup> In that context, it is of interest that a survey of United States paediatricians<sup>20</sup> showed that it will not necessarily prompt a referral to a speech-language pathologist if a parent reports that a child "may be exhibiting signs of stuttering."

On balance, it seems reasonable to state that parents generally know that their children have begun to stutter when they bring them to clinics. Sometimes doctors, or staff at pre-school day care centres or kindergartens, identify children who stutter and prompt parents to bring them to clinics. The clinical task of verifying the presence of stuttering is not challenging.

#### *Stuttering identification across languages*

There is evidence that Dutch speakers can recognise stuttering in Brazilian and Portuguese speakers.<sup>21</sup> In another study,<sup>22</sup> English-Spanish bilingual and English-speaking monolingual observers, who were speech-language pathologists, identified more "disfluencies" in Spanish video speech samples than English samples by the same speaker. It is not clear in this report, but the speaker appears to have been a nonstuttering adult. A study of 18 nonstuttering Spanish-English bilingual 5- and 6-year-olds<sup>23</sup> produced a consistent diagnostic result based on audio recordings of their narratives. More disfluencies were identified that are normally associated with English speaking children of that age.

Another report<sup>24</sup> involved video recordings of readings by two Spanish-English bilingual participants who stuttered and English-speaking speech-language pathologists. For one of the participants, but not the other, the speech-language pathologists noted a higher frequency of stuttering in Spanish than English. However, overall, the authors interpreted findings to "suggest that SLPs [*speech-language pathologists*] can accurately assess and diagnose stuttering in clients from culturally and linguistically diverse backgrounds" (p. 40).<sup>24</sup> Another report<sup>25</sup> involved two Spanish-English bilingual 6-year-olds, one who stuttered and a control who did not. Based on audio recordings, many Spanish-English bilingual speech-language pathologists diagnosed the control child as stuttering. The authors concluded, "it appears that bilingual speakers may be at unique risk for false-positive identification of stuttering" (p. 72).<sup>24</sup> However, although one of those authors has speculated about diagnostic issues with Spanish children with early stuttering,<sup>26</sup> there are no convincing data about the matter as yet.

## Screening for early childhood stuttering

### *The benefits of early childhood stuttering screening*

As with any early childhood health problem, there would be benefits of population screening to identify stuttering as soon as possible after onset. In principle, that screening would allow cases to be identified for clinical management at an optimal period during early development of stuttering.

### *Screening sensitivity and specificity*

Screening is not assessment. It is typically a much briefer procedure than assessment, and it is designed to determine who should have an assessment and who should not. *Sensitivity*, also known as the *true positive rate*, is the proportion of cases correctly identified as stuttering at screening, according to the results of a full assessment. *Specificity*, also known as the *true negative rate*, is the proportion of cases correctly identified as not stuttering at screening, according to the results of a full assessment.

Errors are inherent with any screening process, false negative identification being the most serious of them. This occurs when a stuttering child is erroneously identified as not having stuttering at screening; in other words, when a child really is stuttering but is not identified as such. The clinical issue here is that when it finally becomes apparent that an error has occurred, and that the child really is stuttering, the optimal time for stuttering treatment may have already passed.

False positive identification occurs when a child is erroneously identified as stuttering at screening. In other words, when a child is not really stuttering but is identified to be stuttering. Such an error is unlikely to do any harm because the mistake would become apparent as soon as an attempt at treatment began.

To return to the issue of how accurate parents are at identifying early stuttering when they bring children to clinics, the matter can be restated in the following way. Parents can be thought of as a screening procedure: there is good reason to believe that parent identification of early childhood stuttering is sensitive, with a high true positive rate. However, the specificity of parent identification of early childhood stuttering is unknown. In other words, the false negative rate is unknown.

### *There is currently no accepted screening method*

Much as it is needed, at present there is no generally accepted way to screen for stuttering during early childhood.<sup>27</sup> Surprisingly, such an important topic has attracted almost no research. There was a preliminary report from more than 20 years ago.<sup>28</sup> A more recent, small-scale report involved 36 Lebanese speech-language pathologists, two child stutterers, and six typically developing children.<sup>29</sup> Speech samples were recorded monologues spoken in Lebanese by the children, of between 186–410 words. Results indicated that, regardless of whether the speech-language pathologists used audio or audio-visual samples of the children, this was not a satisfactory method of screening for early stuttering.

## Speech and language disorder comorbidity

### *An ambiguous literature*

Some research has reported how many children have stuttering and another speech or language condition, or have stuttering comorbidities, to use the correct term. There is no doubt that this will occur sometimes.<sup>30</sup> A study of clinician caseloads<sup>31</sup> indicated that 44% of 467 school-age children who stuttered reportedly also had a language or phonological disorder. Another study<sup>32</sup> reported that 34% had articulation disorders and 14% had phonological disorders. However, another report<sup>33</sup> found no such differences between stuttering and control children. A recent report<sup>34</sup> studied 58 stuttering children and 40 control children for a 4–5 year period. No systematic differences were reported for phonology across the period of study. A recent review of the literature<sup>35</sup> concluded that research about the topic is ambiguous. Given such ambiguity in the literature, it is not surprising that clinicians may be uncertain about concurrent management of children with comorbid stuttering and speech sound disorder.<sup>36</sup>

The same ambiguity pertains to findings about language problems with pre-school and school-age<sup>†</sup> children who stutter. Many reports have found that children who stutter have language less advanced than peers<sup>37</sup> and many have found that not to be the case.<sup>38</sup> The 4–5 year study referred to previously<sup>34</sup> found that the child stutterers “though within normal limits” were “slightly behind ... on broad measures of language development” (p. 23). The ambiguity of this literature is highlighted by two reviews of the available literature at around the same time that came to opposite conclusions. One report<sup>39</sup> was a meta-analysis\* of 22 studies with a conclusion that stuttering was associated with lower language test scores than control children. However, two more recent reviews of the literature<sup>40,41</sup> reported that available research did not support any such conclusion. At present, little is known about bilingualism and childhood stuttering.<sup>42</sup> A review<sup>43</sup> of an often-cited seminal study from early last century,<sup>44</sup> which purported to implicate bilingualism in stuttering onset, demonstrated that its results were not believable.

To make the literature even more difficult to interpret, some authors<sup>45</sup> pointed out that no study of language and early childhood stuttering had used “conversational language samples collected in a naturalistic, non-contrived play environment with peers” (p. 649). They developed a method to rectify that situation and showed that it was viable with four children with stuttering in pre-school play environments.

#### *Possible bias*

An issue here is that children who have comorbid speech and language disorders with stuttering are more likely to be referred to a clinic than children who stutter but have no other speech and language disorders. Therefore, published figures could well be overestimates of stuttering comorbidity for stuttering in general.<sup>46</sup> Also, as considered during Lecture Ten, children who stutter could be socially withdrawn. This could be another source of bias because they may be reluctant to speak during language testing, leading to underestimation of their language skills.<sup>38</sup>

### **ADHD comorbidity**

#### *An even more ambiguous literature*

There have been some suggestions during the past two decades of an association between attention deficit hyperactivity disorder (ADHD) and childhood stuttering. In 2003, a tutorial about ADHD and stuttering<sup>47</sup> was based on two research findings at that time which do not seem at all remarkable. The first<sup>48</sup> was a survey of 241 speech-language pathologists about 3–20-year-olds they were treating for stuttering. Their reports suggested that 3% of their caseloads may have had ADHD, which was well within community prevalence rates. The second paper<sup>49</sup> in the 2003 review was a report of 50 child stutterers whose parents used a survey to establish that 26% of them could be described with the vague term “attending disorder.”

Subsequent reports have not done much to clarify this confusing picture, because they have not incorporated generally accepted diagnostic procedures for ADHD. Instead, they report about children with and without “ADHD symptoms” based on parent screening methods,<sup>50,51,52</sup> teacher reports,<sup>53</sup> or adult recall of childhood symptoms.<sup>54</sup> A report of 356 adults<sup>55</sup> found that they were likely to indicate experience of ADHD symptoms, but no diagnoses were included in the report. A report of 3–17-year-olds from the United States National Health Interview Survey (N=62,450)<sup>56</sup> involved children whose caregivers reported stuttering during the previous 12 months, and reported a doctor or health professional having diagnosed ADHD. The boys in the sample had greater odds than girls for a diagnosis of ADHD.

Another report,<sup>57</sup> used 84 adults with a confirmed ADHD diagnosis and 207 controls and found stuttering in 18% of the former group. A problem with this report, however, is that methods to identify

<sup>†</sup> The term school-age refers to children who are at the stage of education commonly referred to as primary school or elementary school, spanning the age range 7–12 years.

\* Meta-analysis is a systematic review that synthesises evidence from numerous empirical reports.

stuttering were not specified. Regardless, 2% of the control group were reported as stuttering, which seems reasonably accurate, as will be discussed later during this lecture.

On balance, all this research is worth noting for two reasons. First, ADHD involves impaired regulation of attention and behaviour, which is a topic that features in research about early childhood temperament and stuttering, which will be considered during Lecture Eleven. Second, the topic is clinically pertinent because, as will be discussed during Lectures Six and Seven, many treatment methods for children require considerable compliance from them, and this might be challenging for children with ADHD. Indeed, one of the reports just mentioned<sup>52</sup> found that children with early stuttering required more time to complete stuttering treatment if they had more “ADHD symptoms.”

### Autism spectrum disorders comorbidity

A review of the topic<sup>58</sup> drew attention to some reports suggesting that this comorbidity might occur.<sup>32,59,60</sup> The authors of the review<sup>58</sup> discussed some features of the speech and language characteristics of children with a comorbid diagnosis of stuttering and autism spectrum disorders, and some views about the assessment and treatment that should occur. They presented two case studies of children with such comorbid diagnoses. There are other case reports of adults and children with such diagnoses.<sup>61,62</sup>

### Legal stuttering identification

Clinicians may be asked to provide a written report to a lawyer or give verbal evidence in a court about a client with stuttering. They might be asked to comment on a claim that stuttering began after a physically or psychologically traumatic event. Or they may be asked to comment on a case of suspected malingering. For example, people who have been heard to not stutter while committing a crime have been known to fake stuttering to give the impression that they could not have been the offender.<sup>63</sup> Publications are available to assist clinicians with preparing such legal assessments.<sup>63,64,65</sup>

### Theoretical perspectives about stuttering identification

#### *The Continuity Hypothesis*

The previous assertion that stuttering identification is not a clinical challenge is based on the idea that stuttering and typical disfluency are categorical things; they are different and, hence, for the most part easily recognisable. But a different perspective about this emerged in 1970<sup>66</sup> in the form of what is known as the Continuity Hypothesis. In effect, this idea is that stuttering and typical disfluency are not categorical things but lie on either ends of a continuum. In other words, stuttering is an extreme form of typical disfluency:

there are few if any aspects of early stuttering which cannot be found occasionally and mildly in the speech of most normal young children. Seen from this point of view, stuttering as a clinical disorder is largely a more extreme degree of certain forms of normal disfluency. (p. 30)<sup>66</sup>

This proposition proved to be controversial, with an experiment shortly after purporting to show that it was wrong,<sup>67</sup> and that stuttering and typical disfluency in fact were “two reliable and unambiguous response classes” (p. 691). There was disagreement about the experiment,<sup>68,69</sup> and some years later another experiment came to the opposite conclusion.<sup>70</sup>

### Conditions to distinguish from stuttering

There are some conditions that have some resemblance to stuttering. Identifying them is straightforward with a case history and basic clinical observation. Their case histories and clinical features are obviously different from stuttering described so far during this lecture, which is sometimes referred to with the term *developmental stuttering* to distinguish it from conditions that will now be reviewed. The first task for a clinician when meeting new clients is to confirm the diagnosis of developmental stuttering rather than one of the following conditions, and to check that stuttering is not comorbid with one of them.

### *Acquired stuttering*

Acquired stuttering is “a broad term and probably the most common one to denote a fluency disorder of non-developmental origin” (p. 42).<sup>71</sup> This guide to terminology for acquired stuttering<sup>71</sup> suggests three terms for subcategories of acquired stuttering. *Neurogenic stuttering* refers to cases arising from neurological damage such as traumatic brain injury, stroke, and neurodegenerative disease. *Drug-induced stuttering* refers to the effects of medication. A detailed review of this topic is available,<sup>72</sup> indicating that 57% of reported cases are linked to antipsychotic drugs. *Psychogenic stuttering* refers to “a dysfluency that is somehow associated with a psychological problem or an emotional trauma” (p. 42).<sup>71</sup>

Reports about psychogenic stuttering are common and clinically puzzling, and it is possible that some or all reported cases are a combination of neurogenic and psychogenic factors.<sup>73</sup> The most recent report about this matter<sup>74</sup> suggests a differential diagnostic procedure for neurogenic and psychogenic stuttering, and it suggests assessment and treatment procedures. The report presents two case histories of mild traumatic brain injury caused during military action.

Neurogenic stuttering seems to be more common than drug-induced stuttering and psychogenic stuttering. Reviews of neurogenic stuttering are available.<sup>75</sup> It appears that those affected by neurogenic stuttering are rarely anxious about it, which is the exact opposite of developmental stuttering, as will be considered during Lecture Ten. Nonverbal superfluous behaviours are common with developmental stuttering but seem to be rare with neurogenic stuttering. One report,<sup>76</sup> based on five cases of neurogenic stuttering and 35 cases of developmental stuttering, suggests that “phonetic, word class, word length, and word position variables” (p. 1) are more similar than different for the two conditions. Another report,<sup>77</sup> with 3-minute video samples of four cases of neurogenic stuttering and four cases of developmental stuttering, suggests that the difference between the two may not be easy to distinguish. On balance, then, it seems essential during assessments for clinicians to explore the client case history to exclude any chance of neurogenic stuttering being mistaken for developmental stuttering. One report<sup>78</sup> described stuttering onset after concussion in three adolescents and two 2-year-olds who had no family history of stuttering. Contrary to usual cases of developmental stuttering, the three adolescents recovered within 8–10 weeks. An audio recording is available for an 18-year old with stuttering after concussion.<sup>79</sup> Neurogenic stuttering has been reported to occur after long COVID-19 infection.<sup>80</sup> There is a case report of a 62-year-old who presented to an emergency department with episodes of stuttering, and who was shown to have had a small infarct.<sup>81</sup> There is a report<sup>82</sup> of an adult presenting to an emergency department with rapid onset of initial-syllable repetition, which resolved two hours later. MRI showed “a small acute ischemic stroke in the left frontal cortex”(p 2). The authors suggested “considering even minor strokes in the differential diagnosis of stuttering” (p. 1).

Overall, neurogenic stuttering is a poorly understood condition, but understanding of it has improved with a study of 319 hospital patients with a mean age of 71 years.<sup>83</sup> Of that group, the researchers diagnosed 5.3% with neurogenic stuttering, with 2.5% of the group having the condition for 6 months. There was considerable comorbidity among the 17 patients diagnosed with neurogenic stuttering. Eleven of them also had aphasia, nine had dysarthria, two had apraxia, and five had cognitive problems. Symptoms described as “stuttering” are sometimes reported after concussion. For example, one report<sup>84</sup> stated “difficulty initiating speech, often repeating ‘dadadada’ before finding her words” (p. 137). A detailed case report is available.<sup>85</sup>

A report analysed data from 20 published reports of neurogenic stuttering after stroke, a cohort of 20 adults with neurogenic stuttering after stroke, and 20 adults with persistent developmental stuttering.<sup>86</sup> The authors concluded

... that lesions causing acquired neurogenic stuttering map to a common brain network, centred to the left putamen, claustrum and amygdalostratial transition area. The association of this lesion-based network with symptom severity in developmental stuttering suggests a shared neuroanatomy across aetiologies. (p. 2203)

### *Cluttering*

It is possible that someone who has *cluttering*<sup>87,88,89</sup> could be mistaken for having stuttering. A study of two Turkish speech-language pathologists showed that they could not subjectively distinguish between the two conditions.<sup>90</sup> The prevalence of cluttering has not been established definitively. A survey report of 1,582 university undergraduates in Israel,<sup>91</sup> found that 23% of them thought that they cluttered after reading a description of it, and that 3.5% reported being treated by a speech-language pathologist for cluttering. The features of cluttering<sup>87</sup> are rapid and mostly irregular articulation, disfluencies that are dissimilar to those of stuttering, and impaired intelligibility because of indistinct and abbreviated articulation. Stuttering and cluttering can be comorbid, with a recent report showing seven of 11 participants with cluttering to also have stuttering.<sup>92</sup> So, as well as someone with cluttering being mistakenly identified as stuttering, it is possible for someone to have both conditions and for stuttering to be overlooked. The World Health Organization defines cluttering as follows:<sup>93</sup>

A rapid rate of speech with breakdown in fluency, but no repetitions or hesitations, of a severity to give rise to diminished speech intelligibility. Speech is erratic and dysrhythmic, with rapid jerky spurts that usually involve faulty phrasing patterns.

### *Tic syndromes of early childhood*

It is possible, but rare, for diagnostic confusion to occur with stuttering and childhood tic disorders, many of which are transient during childhood.<sup>94</sup> Such confusion is most likely to occur when tics have a vocal component. Motor tics (nonverbal tics) will occur when people are not speaking, and that does not happen with the superfluous behaviours of stuttering. The most likely error is for Tourette Syndrome to be mistaken for stuttering. Tourette Syndrome requires one or more vocal tics and two or more motor tics for diagnosis. A report suggested that as many as one fifth of children with Tourette Syndrome may have speech that resembles stuttering.<sup>95</sup> A review of 122 studies is available, which explored “similarities and differences between stuttering and tics in terms of epidemiology, comorbidities, phenomenology, evolution, physiopathology, and treatment” (p. 1).<sup>96</sup>

### *Neurological disorder*

When extremely severe cases of stuttering develop suddenly during early childhood, it may be mistaken for a neurological disease. Doctors have been known to refer cases of severe early childhood stuttering for neurological evaluation. However, speech-language pathologists usually don't make that mistake. On that topic, there is an interesting report of three adult sisters who were diagnosed with late-onset Tay-Sachs disease.<sup>97</sup> The report states that the first of them “developed a stutter at approximately age 10” and the second “developed a stutter at age 8” (p. 289). Videos of the participants accompany the report, and the second participant states during the video that she stuttered as a child. The videos clearly show speech motor problems, but a diagnosis of developmental stuttering is not warranted for any of the participants. Similar diagnostic issues were present in another report involving 453 patients who received deep brain stimulation treatment for Parkinson's disease and who were reported to be stuttering afterward.<sup>98</sup> A report of 100 Parkinson's disease patients and 25 controls<sup>99</sup> reported “twice as many stuttered disfluencies during conversation compared to control participants” (p. 1).

### **An unusual case history<sup>†</sup>**

The following case history illustrates a rare instance of when someone presents to a speech clinic with stuttering but obviously it is not straightforward developmental stuttering. Potentially, this stutterer is comorbid with one or more of the conditions that were just described.

A 9-year-old boy presented to a clinic with no family history of stuttering or reports of him or his twin brother ever stuttering. He recalled that while camping with family and friends he showered after swimming and then noticed that he was stuttering. Shortly after, at assessment, the clinician noted

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<sup>†</sup> Thanks to Michelle Taylor for this case history.



syllable repetition, incomplete syllable repetition, and nonverbal superfluous behaviours of muscle contractions around his mouth.

Two weeks later the stuttering stopped, and the clinician did not hear from the family again until 18 months later, when his mother reported that the stuttering had returned. The clinician saw the boy again and observed tics as well as stuttering, and so suggested assessment by a paediatric neurologist. The clinician also suggested a psychiatric assessment, and his mother was receptive to that idea, having been concerned about her son's anger and sensitivity.

## STUTTERING AND GENETICS

### Background

It has been known for a long time that genetics is involved with stuttering. For a speech-language pathology readership, comprehensive<sup>100,101</sup> and compact reviews<sup>102</sup> are available, along with an overview of the current status of the field.<sup>103</sup> More technical reviews are available<sup>104,105,106</sup> with more focus on the science of genetics, and there is an overview of specific genetic mutations identified to date.<sup>107</sup> A review paper<sup>100</sup> outlines the progress in accumulating knowledge about genetics and stuttering in four methodological phases: familial incidence, twin studies, family aggregation, and biological genetics. The subsequent overview follows those headings.

### Familial incidence

#### *Family history is common*

Fundamental evidence for genetic involvement with a condition is vertical transmission: in other words, a family history. A review of 25 reports dating from 1937 to 2019<sup>101</sup> (Table 2-5, p. 65) shows that, overall, around two-thirds of those who stutter report a family history. So, it is more likely than not that a client will report a family history.

#### *Underestimates are likely*

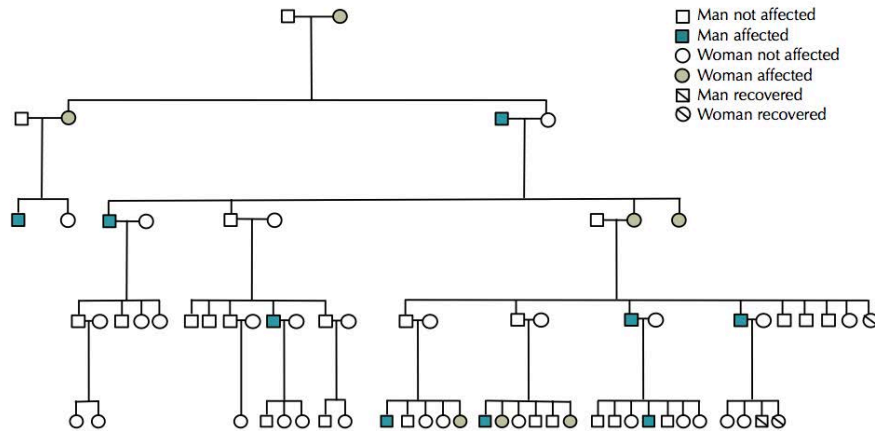
There is good reason to believe that such participant-report data underestimate the true family history rate, with many family members having affected relatives but under-reporting them.<sup>108</sup> So, clinically, if an interview suggests that there is no family history, there is some room for doubt. For example, the informant may not have much contact with all relatives or may simply have forgotten about a relative who stutters.

#### *A famous family*

The diagram\* is from a 1940 report of five generations of an Iowa family in the United States.<sup>109</sup> During clinical practice clinicians routinely see many families with stuttering occurring through them, although not usually as densely as this example. It is probably worth learning to draw family pedigrees, or learning to use a program that constructs a pedigree, for noting in clinical files. The symbols in the diagram (but not the colour coding) are standard.

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\* Adapted and reproduced with permission: Gray, M. (1940). The X family: A clinical and laboratory study of a "stuttering" family. *Journal of Speech Disorders*, 5, 343–348. © 1940 American Speech-Language-Hearing Association.



### Males are affected more than females

Proportions of boys at reported onset range from 50%,<sup>207</sup> 61%,<sup>204</sup> and 68%.<sup>212</sup> The ratio of men to women affected by stuttering ranges from 3:1 to 5:1 in various reports,<sup>101</sup> which translates to 75–83% men. There are two features of those figures to note. First, there are more males than females who stutter. Second, considering all publications about the matter, the reported ratios for young children seem to be more evenly balanced between genders than are ratios for adulthood. So, it seems that girls are more prone to recovery than boys. There have been independently replicated reports linking the gender imbalance to prenatal testosterone levels,<sup>110,111,112</sup> as occurs with some other conditions.

### Studies of twins

#### *Identical and non-identical twins*

Studies of twins are another way of establishing a genetic basis to a condition. Identical twins (monozygotic) are genetically identical people, but non-identical twins (dizygotic) are like any other siblings, except they develop *in utero* at the same time. If both twins have a condition it is referred to as concordance, and if only one twin has a condition it is referred to as discordance.

#### *Monozygotic concordance is greater than dizygotic concordance*

If the incidence of monozygotic concordance for stuttering is higher than dizygotic concordance, it strongly suggests that a genetic factor is involved. In other words, if the incidence of identical twins both stuttering is more than that for non-identical twins, then it suggests genetic involvement.<sup>†</sup>

It seems to have been discovered in the 1930s that, indeed, monozygotic concordance for stuttering is greater than dizygotic concordance,<sup>113,114</sup> with the first attempt to quantify concordance rates some years later.<sup>115</sup> In 1981 a seminal study of 30 twins<sup>116</sup> was published and its findings were subsequently replicated with larger cohorts.<sup>117</sup>

A large parent self-report study of 10,500 5-year olds<sup>118</sup> found, for reports of “probable stuttering,” dizygotic concordance for boys of 36% and 34% for girls, and monozygotic concordance for boys of 53% and 61% for girls. This report estimated “probable stuttering” to be 42% inheritable; in other words, a 42% genetic contribution to stuttering. The highest estimate of genetic contribution to stuttering occurred in a self-report study<sup>119</sup> of 1,896 11-year-old twin pairs, which suggested 80% and 85% genetic contributions to stuttering for boys and girls, respectively.

In short, both twins who are identical are more likely to stutter than are non-identical twins. But monozygotic concordance for stuttering is not 100%. A broad interpretation of these studies is that

<sup>†</sup> The assumption underlying this reasoning is that the living environments of both types of twins are the same.

much—or perhaps the majority—of stuttering can be accounted for by genetic factors. The remainder of cases would be accounted for by genetic or nongenetic factors that are not known about yet.

### Family aggregation studies

#### *Clues about genetic models*

Statistical analyses of family history data can give some clues about how a condition is transmitted genetically.<sup>100</sup> However, the retrospective methods of that approach, relying on recall, are a limitation of the method. It appears that two reports during the 1930s<sup>120,121</sup> were the first applications of the family aggregation method with stuttering. Following this, after two and a half decades, was the first comprehensive report.<sup>122</sup> This study involved 213 people with stuttering, or probands to use the proper genetic term, and their families. This study showed an increased stuttering incidence among first-degree relatives of probands—parents, siblings, and children—than for the general population. The report also found that fathers and brothers of those affected had more than twice the stuttering incidence as mothers and sisters of those affected.

#### *Take-home messages*

Kenneth Kidd and colleagues at Yale University added to the family aggregation data,<sup>123</sup> and in the early 1980s they published data about 600 probands who attended clinics.<sup>124,125</sup> A take-home message was provided by pooling data for families covered in the first comprehensive report<sup>122</sup> and the Kidd studies:

For men who ever stuttered, 9% of their daughters and 22% of their sons will be stutterers; while for the fewer women who ever stuttered the risks are higher, as 17% of their daughters and 36% of their sons will be affected. (p. 229)<sup>126</sup>

This information implies that although women are affected by stuttering less often than men, they are more likely to have a child who stutters.

A more recent publication<sup>127</sup> involved 739 probands, of which 60% reported a family history. Three-quarters were male. There were 431 probands 5 years or younger, 59 were 6–12 years, 97 were 13–17 years, and 152 were 18 years or older. Stuttering severity was established with parent-reported or self-reported rating scales. The impact of stuttering was measured with the Overall Assessment of the Speaker's Experience of Stuttering (OASES) (see Lecture Four). Key findings were:

- (1) All probands had 2–3 times greater odds of having a father than a mother who stuttered
- (2) Two thirds of cases experienced moderate impact of stuttering
- (3) A fifth of cases experienced moderate-severe or severe impact of stuttering
- (4) Family history and sex did not appear to influence stuttering severity
- (5) Boys have stuttering onset 5.5 months later than girls; 9 months when there is no family history
- (6) For those with a family history, impact of stuttering was 2.5 times greater for females than males.

Those results are consistent with OASES scores for 162 adult stutterers, ages 7–30 years.<sup>128</sup> The bulk of participants were in the mild-moderate and moderate impact range of scores, with females impacted overall more than males. The impact of stuttering was greatest during the adolescent years.

#### *Stuttering and birth rank*

Sometimes when there is a family history of stuttering, parents can be concerned that stuttering could be transmitted by non-genetic means, such as with a sibling “copying” the stuttering of another sibling. Parents who stutter might have a similar concern that somehow their stuttering will encourage their children to begin stuttering.

A report from the Yale group<sup>129</sup> verified that such concerns are not justified by showing no association between birth rank and stuttering among siblings. If stuttering could be transmitted by sibling “copying,” children born earlier in the family would have more opportunities to copy stuttering models than children born later in the family, and a different finding would have been expected from the Yale data. Consequently, there is no reason for parents to be concerned that stuttering can be

transmitted by “copying.”

## Biological genetic evidence

### *Genome-wide studies*

Direct genetic evidence is another technique to explore the genetics of stuttering. Until recently, the most up-to-date procedure was with genome-wide linkage studies, which trace patterns of stuttering inheritance through generations using genetic markers. The review paper mentioned earlier<sup>102</sup> lists six studies that have provided evidence of multiple chromosome linkage.<sup>130</sup> These studies make it seem probable that stuttering is polygenic in nature, meaning that many genes are involved in the genotype, and raising the likelihood that other genes are involved and await discovery. However, it has been noted that there is little consistency of results across studies reported to date<sup>100</sup> (see Table 3, p. 42). The genome-wide association study, or whole-genome association study, is a technique that involves those affected and not affected with a condition. To date, this method has been reported in one study about stuttering, which again suggested polygenic inheritance.<sup>131</sup>

### *Candidate gene analysis*

Another source of biological genetic evidence of stuttering is candidate gene analysis, which provides information about contributions of specific genes. Reports using this method have identified mutations in four genes to date (GNPTAB, GNPTG, NAGPA, AP4E1).<sup>132</sup> It is possible that a mutation in these four genes could cumulatively account for 20% of stuttering in unrelated cases.<sup>106</sup>

These gene mutations are part of a biological lysosome cell pathway. Lysosomes are small membrane sacs in animal cells that contain many enzymes and are often described as the cell’s recycling bin. They are responsible for many metabolic functions, and mutations of the genes that encode lysosome enzymes cause many lysosomal storage diseases. It seems that mild mutations of the genes are associated with stuttering, and severe mutations are associated with serious conditions including neurological disease involving intellectual disability and white matter pathology.

Two studies have implicated gene mutations with grey matter volume development and energy metabolism in the development of stuttering.<sup>133,134</sup> The latter of those studies<sup>134</sup> were followed up with a magnetic resonance imaging study of 26 child stutterers and 44 controls, with a mean age 6.5 years. For both groups there was a correlation between glucose uptake and grey matter volume in brain regions involved with speech and language processing. The researchers suggested that these findings could somehow be involved in early childhood stuttering development.

### *A mouse model of stuttering*

Considerable interest has been generated recently with an (arguably adventurous) attempt to establish a mouse model of stuttering.<sup>135</sup> A so-called knock-in mouse was developed with a mutation in the GNPTAB gene, which is associated with the lysosome cell pathway. Compared to control mice, the researchers reported that the ultrasonic vocalisations of the mice with the mutated gene were fewer and with longer pauses between. More repetitions of vocalisations were also reported for the experimental mice. The researchers concluded that their results established “the mouse as an attractive model for studying this disorder” (p. 1009).<sup>135</sup> A subsequent publication<sup>136</sup> reported the same effect with two other gene mutations associated with stuttering, and suggested astrocytes—a central nervous system cell—“as a site of the neuropathology, leading to a deficit in interhemispheric connectivity in stuttering” (p. 17515).

### *Genetic mutations and stuttering treatment*

There has been a preliminary attempt to assess whether carrying a mutation in one of the four genes mentioned earlier is associated with the outcome of stuttering treatment.<sup>137</sup> The authors of that work astutely remark that, although they can account for only a minute proportion of stuttering cases, “these four genes are closely related functionally” and that “all are involved in the process of intracellular trafficking” (p. 12). They argue that “deficits in this cellular function are now recognized as causative of a wide range of neurological disorders” (p. 12). They draw on the results of their knock-in mouse

study<sup>135</sup> to suggest “that the control of vocalization is an innate, conserved biological process, and that genetic deficits in this control could affect therapies designed to correct such deficits” (p. 12). However, assessing the effects of genetic mutation on stuttering treatment outcome is certain to be a complicated and protracted endeavour.

### Applying information about stuttering and genetics to clinical practice

Clients and their parents who don't know it already can be usefully informed that genetics is involved with the condition, rather than it being a psychological problem, as suggested by common stereotypes discussed during Lecture One. This may counteract misinformation from those stereotypes. Clinically, it can also be useful to introduce a discussion of what causes stuttering by stating that genetics is involved with it. That discussion may be usefully combined with information—discussed shortly—about the physical nature of atypical neural processing involved with stuttering. Genetic risks may be particularly useful to discuss in cases where pre-school children are being treated for stuttering and there are younger siblings in a family who are at risk of onset.

### Conclusions

The genetics of stuttering is complex, with incomplete penetrance within families. It is virtually certain that the influence of genetics on stuttering is polygenic, involving combined actions of more than one gene, such as with hypertension and coronary heart disease. Around 70% of those who stutter report a family history, so genetic involvement appears not to be always present, suggesting that genetics is not necessary for stuttering. And studies of twins make it obvious that genetics does not always lead to stuttering, so it is not sufficient for stuttering. Even though studies of twins support estimates that stuttering is around 80% inheritable, genetic mutations have been discovered that can explain no more than 20% of stuttering occurrences, with many of those mutations discovered in stuttering-dense consanguineous families.

## ATYPICAL NEURAL SPEECH PROCESSING<sup>†</sup>

### Overview

There is extensive evidence from neuroimaging research with children and adults who stutter implicating the white fibres that connect areas in the brain that are critical for the complexity of spoken language. This research literature expands each year, and a 2018 edition of the *Journal of Fluency Disorders* was assigned specifically to the topic.<sup>138</sup> Reviews and overviews of neuroimaging studies cite an extensive body of research that is accumulating rapidly.<sup>139</sup>

This research incorporates evidence of unusual structural and functional non-dominant—right sided—brain activity in speech areas. A 2020 review<sup>140</sup> presents 47 papers that implicated functional issues with the supplementary motor area, which is responsible for planning and execution of complex movements. Atypical neural processing associated with stuttering may not be associated specifically with speech functions. One paper reported “wide-spread decreases in connectivity for adults who stutter in regions associated with sensorimotor, cognitive, emotional and memory-related functions.” (p. 1)<sup>141</sup> A 2023 meta-analysis<sup>142</sup> concluded that:

developmental stuttering is characterized by structural and functional alterations in multiple brain networks that support speech fluency or sequential motor processes, including cortico-cortical and subcortical connections. (p. 11)<sup>142</sup>

A review of the topic concluded that

Both children and adults who stutter show atypical brain structure and functional patterns that can be localized and form part of a number of major neural networks. Implicated are cortical areas of the speech motor planning and control networks ... including frontal lobe regions such as the motor cortex,

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<sup>†</sup> Thanks to Ann Packman for guidance with this material.

premotor cortex, inferior frontal gyrus, frontal operculum, insular cortex, and presupplementary and supplementary motor areas ... Also implicated are parietal and temporal perisylvian regions, such as the supramarginal gyrus, and higher order auditory regions, which might be related to differences in sensorimotor integration and feedback control ... Furthermore, subcortical structures such as the basal ganglia, thalamus, and cerebellum are implicated, which may relate to differences in learning, initiation, timing, sequencing, and error monitoring functions ... (p. 3–5)<sup>143</sup>

### Three essential considerations

There are three essential considerations when interpreting the neuroimaging literature. The first is that there is no single brain region responsible for speech and language. Speech and language is a network-level process involving many brain regions. Second, neuroimaging research has been conducted mostly with reference to observable stuttering moments. In other words, it has used an objective definition of stuttering, rather than an internal definition that accounts for the experience of stuttering (see Lecture One).<sup>143,144</sup> The third consideration is that, to a neurologist, the brains of children and adults who stutter look normal. There are no gross abnormalities. Sophisticated instruments and group statistical analyses are needed to detect differences from peers.<sup>143</sup> This third consideration is essential to convey to clients and parents when describing these research findings.

When using current imaging technologies to examine the brain of an individual who stutters, it is unlikely that we will detect apparent morphological or functional anomalies. Both gray and white matter structures will appear well-formed and in their proper places in persons who stutter, and initial observations of brain waves will not appear atypical. Stuttering does not exhibit overt signs like a visible fracture in an X-ray or the clear disorganized electrical activity seen in an electrocardiogram for cardiac arrhythmia. (p. 3).<sup>143</sup>

### White and grey matter

The more recent studies suggest that the issue is one of connectivity; transmission of information along the white matter fibres of the brain are atypical in areas involving spoken language. White matter fibres form complex transmissions of information between executive areas of the cortex and are critical for the development of complex neural networks needed for spoken language. Atypical neural processing is not constrained to white brain matter. There are reports that grey matter of the brain is affected in adults<sup>145</sup> and children.<sup>146,147,148</sup> The contribution of grey matter involves information processing. One report<sup>149</sup> used the innovation of studying spontaneous speech with functional magnetic resonance imaging using a method to control for movement artifacts during speech. With 22 child stutterers and 18 controls, ages 5–12 years, the stuttering group showed decreased activation in the left premotor cortex.

### The arcuate fasciculus and the corpus callosum

Reviews of brain imaging research<sup>139,143 150,151</sup> and subsequent publications,<sup>152</sup> show, compared to controls, differences that have been detected in two prominent white matter brain structures for adults and children who stutter: the corpus callosum and the arcuate fasciculus. However, differences are not constrained to those structures. Differences in other white matter structures have been identified, such as the superior longitudinal fasciculus, frontal aslant tract, cerebellar peduncles, and corticobulbar tracts.<sup>143</sup>

The *corpus callosum* is a large white matter fibre structure connecting the two hemispheres of the brain. The *arcuate fasciculus* is a curved, bilateral bundle of white matter fibres that are fundamental to speech and language production, linking parts of the cortex responsible for expressive and receptive language. These are traditionally known as Broca's area and Wernicke's area, respectively.

An essential context for all this information, however, is that it is not constrained to stuttering. For example, atypical neural processing has been reported to be associated with specific language impairment<sup>153</sup> and speech sound disorder.<sup>154</sup>

## A current hypothesis

Some researchers<sup>155</sup> have drawn on the results of neuroimaging research, and genetic research implicating lysosomal metabolism, to present a testable hypothesis: the onset and development of early childhood stuttering is linked to atypical or late myelination<sup>†</sup> of perisylvian fibre tracts. A publication has sustained this hypothesis.<sup>156</sup>

## The critical issues

### *Atypical neural processing is not sufficient for stuttering*

It is obvious that atypical neural processing, alone, is not sufficient for stuttering to occur. That is obvious because of the many situations in which those who stutter can speak without stuttering, such as the fluency inducing conditions described during Lecture One. Also, as will be discussed shortly, stuttering does not occur when children first start to speak but occurs sometime after during early language development. If atypical neural processing is somehow fundamental to stuttering, something additional must occur for stuttering to appear.

### *Is atypical neural processing necessary for stuttering?*

What is yet to be determined is whether atypical neural processing is necessary for stuttering to occur: whether it is always present when stuttering is present. And even if atypical neural processing is necessary for stuttering, it needs to be determined whether it is part of the cause of stuttering or, as several researchers have suggested, is a consequence of it.<sup>139,152,157</sup> The latter explanation is plausible considering the cortical plasticity of the developing brain,<sup>158</sup> and the fact that not only does the brain drive behaviour but behaviour can change the brain.<sup>159</sup> This issue does not pertain specifically to stuttering. It has, for example, been discussed in relation to developmental language disorders.<sup>160</sup>

Some suggestion—but not conclusive evidence—of a causal role has emerged from findings of grey and white matter structural anomalies for participants with stuttering ages 8–13 years compared to control children,<sup>161</sup> and similar results for younger children 3–10 years old.<sup>146,162,163</sup> This issue has been summarised as follows:

... studying children who stutter (CWS) is important because the brains of CWS have had far less time to change in response to stuttering; observation of differences in brain activity in CWS is, therefore, more likely to reflect causal mechanisms of the disorder. (p. 3).<sup>139</sup>

There has been a report<sup>164</sup> of differences in cerebral blood flow in several brain regions for 35 stuttering 4–14-year olds and 27 matched controls.

### *Evidence for causality*

Lecture Three deals with the cause of stuttering and, in short, there is no convincing evidence yet that atypical neural processing is causal in nature, rather than resulting from years of stuttering. There is only an implication—a substantive one—from its existence in children that it is causally related. Direct evidence would include findings that it is necessary for stuttering to occur, being present in all those who stutter. Researchers have indicated that another source of such evidence would be data from longitudinal studies with age-matched controls.<sup>152</sup> Such evidence has been provided.<sup>155</sup>

Another key source of evidence for causality would be the existence of atypical neural processing prior to stuttering onset in genetically at-risk children.<sup>165</sup> There is some preliminary evidence to that effect, with a study of neonates who were genetically at risk of stuttering, and controls.<sup>166</sup> Those preliminary findings suggested that corpus collosum differences from controls exist shortly after birth. No indications were found for arcuate fasciculus differences. The researchers suggested that the latter finding may have occurred because the fibre tracts in the left hemisphere responsible for speech do

<sup>†</sup> Myelination is an early developmental process that coats each axon of neurones with a fatty substance called myelin. The myelin sheath provides optimal speed and efficiency of nerve cell transmission.

not fully myelinate until the age of 2–3 years.<sup>152</sup>

A large-sample cross-sectional study<sup>167</sup> involved participants in the age range 3–58 years, 166 of whom stuttered, and 191 controls. The study suggested developmental changes across the lifespan, but no consistent differences from controls that applied to both children and adults. The study used surface-based measures of cortical thickness and gyrification (folding of surface gyri and sulci). Compared to matched controls, the children who stuttered had a thinner cortex in certain speech-related regions. The researchers suggested that this indicated differences in developmental trajectory. For the children there was a significant correlation between cortical thickness and stuttering severity. The adults who stuttered, compared to controls, showed “reduced gyrification in the right inferior frontal gyrus” (p. 1). The researchers noted that genetics strongly influences cortical development, and hence suggested that genetics may influence differences in developmental brain trajectories between children who stutter and controls. They also suggested that the different finding for children and adults may reflect the experience of persistent stuttering. In other words, compared to children, experiences and compensations for stuttering with adults may have a more pronounced and variable effect on brain morphology, making it difficult to find significant group differences.<sup>†</sup>

### Clinical applications of neuroimaging research

At present, treatments based on neuroscience research are not available, but this is likely to eventually occur.<sup>144</sup> Ideally, the benefits of research about atypical neural processing with stuttering will eventually improve treatment for it.<sup>168</sup> In the interim, the authors of that article note that the clinical implications of this research so far are that those who stutter “will be buoyed to know that the myth of stuttering as a psychological/psychiatric disorder is being debunked by current research illuminating the neurological foundations of stuttered speech” (p. 116). Other authors have expressed a similar view:

Regardless, at present, neuroscience does have practical clinical application. The reality of neuroplastic change is motivational for clients and parents. Neuroscience conveys the physical nature of stuttering to them, and that they are to be free of blame for it. Also, a neuroscience explanation of stuttering can alleviate negative emotions about stuttering and associated self-stigma. (p. 6)<sup>144</sup>

Indeed, this research may be eventually destined to alleviate the social marginalisation of those affected by stereotypes, as discussed earlier during this lecture. It should also contribute to alleviating the lasting impact of decades of theorising about stuttering being a psychological problem, as discussed during the next lecture.

## EPIDEMIOLOGY OF STUTTERING

### Epidemiology

Epidemiology has come to international prominence with the Covid pandemic. It is the study of health issues and problems in populations, and factors that influence them. There are two types of epidemiological research designs: *observational* and *experimental*. Observational research designs do not attempt any treatment, and common methods of these are cross-sectional studies, cohort studies, or case-control studies. Common experimental studies that test treatments in populations are randomised controlled trials or quasi-experimental designs.

In the case of stuttering, the favoured methods are observational; it is not ethical to do experiments that expose children to things that might cause stuttering. The only published example of such an experiment in stuttering research is infamous,<sup>169,170</sup> with eventual consequences of a public apology from the university concerned and legal compensation to the participants decades later.

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<sup>†</sup> Thanks to Soo-Eun Chang for assistance with this paragraph.



## The value of stuttering epidemiology

Stuttering epidemiology research can provide useful information for day-to-day clinical practice. Perhaps most importantly, it can establish how prevalent stuttering is and provide information about its natural developmental course through early childhood if it is not treated.

Epidemiological studies can compare children who begin to stutter with those who do not. Such studies can provide clues about what might cause, or somehow be associated with, stuttering onset and development. Such research can also provide clues about how to predict which children will begin to stutter.

## Epidemiology and public health

Apart from day to day clinical practice, epidemiological information has a broader impact on stuttering treatment services because it establishes public health information that can change government health care policy. In cases where a condition occurs frequently, causes significant distress, and can be successfully treated—as is the case with stuttering—information to that effect can prompt governments to provide adequate health care services for it. In cases where adequate health care services are lacking, that situation can be repaired by astute advocacy from clinicians, those who stutter, and the public. There are examples of public advocacy leading to Government enquiries and reports about communication disorders, which have included stuttering.<sup>171,172,173</sup>

## POINT PREVALENCE OF STUTTERING

### Point prevalence

*Point prevalence* of a health issue or condition, often referred to simply as prevalence, is how many people are affected by it at any one time. The most common method of establishing point prevalence for a condition is to use a cross-sectional design, where a population sample is assessed at one time. Often, prevalence studies involve assessments at different ages.

### Two essential caveats

A comprehensive review of stuttering epidemiology research up to 2012<sup>102</sup> details all the caveats that need to be kept in mind about stuttering prevalence. However, there are two central caveats that have the overall effect that estimates of stuttering point prevalence could well underestimate the true value.

#### *Identifying participants*

In the case of stuttering, ideally, researchers would assess all participants in a study to determine whether they are diagnosed with stuttering or not. But for practical reasons more than anything else, commonly that does not happen, and most of the available stuttering point prevalence information comes from reports given by relatives, or by self-report.

As discussed during the previous lecture, although self-report about stuttering may be believable for those presenting to clinics for help, the same may not necessarily apply to those who are recruited from the general population to participate in a study. For example, many adults will not necessarily recall having periods of stuttering when they were children, and they may not recall such childhood experiences of their relatives. Yet a common method with cross-sectional study of stuttering populations is to ask those who stutter or their relatives about recall of stuttering within their families. This is known to be a notoriously unreliable procedure, with one report finding that it results in overestimates of stuttering history within families,<sup>174</sup> and another finding the opposite.<sup>175</sup>

#### *A socially avoidant population*

Another potential problem is that those who stutter quite often will, to some extent, be socially avoidant because they are socially anxious, as will be discussed during Lecture Ten. So, because a point prevalence study of stuttering requires a one-off social engagement of participants with researchers who are strangers, point prevalence estimates of stuttering could well underestimate the

true value.

### Estimates of stuttering point prevalence

A well-known reference text<sup>101</sup> documents 46 international prevalence studies from 1893 to 2019 dealing with children (Tables 2-1 and 2-2, p. 43–45). The mean reported point prevalence in those tables is 1.6%. A recent estimate<sup>176</sup> was 1.7%, based on 3,437 children 6–15 years old in India. However, the standard deviation in reports is quite large at 1.9, because the 46 estimated values vary considerably. The lowest reported prevalence figure is 0.3% and the highest is 11.2%. It is of interest that the data for United States children (Table 2–1) have a mean of 1.1% prevalence with standard deviation of 0.6, but the data for other countries (Table 2–2) have a mean of 2.0%, with much more variability, having a standard deviation of 2.3. This suggests that either the point prevalence of stuttering varies internationally, or that the variation is some kind of statistical error arising somehow from different methods used in the two sources.

The latter seems to be the most likely explanation, since there is no sound theoretical reason to suppose that the point prevalence of stuttering would vary so much from country to country. In fact, it has long been accepted that stuttering prevalence is the same for all races and languages.<sup>177</sup> When a speaker is bilingual, stuttering occurs in both languages.<sup>178</sup> However, there are reports that more stuttering is likely to occur with a second language than a first.<sup>179</sup> A review<sup>180</sup> highlighted that nearly half the world population is bilingual or multilingual, yet surprisingly little is known about how this relates to stuttering. Those authors conducted a systematic review that found language proficiency and dominance to influence stuttering severity. However, they found little support for the idea that syntactical and phonological differences between languages, and their syllable structure, are involved in determining stuttering severity in bilingualism. An account of the historical origins of stuttering and bilingualism is available.<sup>181 †</sup>

The review article mentioned previously<sup>102</sup> notes that one large-cohort study (N=119,367)<sup>182</sup> reported more stuttering among African Americans than other Americans. Why that could be the case is challenging to explain, as is the convincingly reported high prevalence among those with Down Syndrome. A review of the pertinent literature<sup>183</sup> drew attention to reports that 10–45% of those with Down Syndrome stutter. A later report of children with Down Syndrome 3–13 years old estimated a 30% prevalence.<sup>184</sup> The most recent systematic review<sup>185</sup> found 14 pertinent reports, with 1,833 participants, with an age range 3–58 years. Prevalence was reported in the range 2.4–56%.

A review article<sup>102</sup> presented a table of prevalence studies conducted this century, along with the conclusion “it is clear that prevalence under age 6 is considerably higher than in later periods in life” (p. 74). A more detailed version of that table is presented below.\* As the authors of the review article note, their conclusion is consistent with the occurrence of natural recovery after onset, as will be discussed shortly. Their conclusion is also consistent with early childhood stuttering being particularly responsive to treatment compared to treatment in later periods of life, as discussed in subsequent lectures. Also, as the authors note, it may well be the case that early childhood stuttering treatment interacts positively with a trend for natural recovery. Other authors have also offered this suggestion.<sup>186</sup>

PARTICIPANTS	AGE IN YEARS	PREVALENCE	MALE/FEMALE RATIO	
1,113 <sup>187</sup>	4–5*	2.2	0.7:1	* Not clear in the report but this probably is the age range
4,983 <sup>188</sup>	4.5	5.6	**	** Not reported
3,165 <sup>189</sup>	2–5	2.6	2.6:1	
10,000 <sup>190</sup>	5–13	0.3	**	

† Copies of this Bulgarian publication are available from the author on request: john.vanborsel@ugent.be

\* Adapted and reproduced with permission: Yairi, E., & Ambrose, N. (2013). Epidemiology of stuttering: 21st century advances. *Journal of Fluency Disorders*, 38, 66–87. © 2013 Elsevier.

21,027 <sup>191</sup>	6–10	0.8	5.1:1
	11–15	0.5	4.7:1
	16–20	0.3	1.9:1
12,131 <sup>108</sup>	2–99	0.7	2.3
	3–10	2.0	2.5
119,367 <sup>182</sup>	3–10	2.0	2.5
	11–17	1.2	

The most recent calculation of adult stuttering prevalence<sup>192</sup> was based on a review indicating that two reports<sup>108,193</sup> (the latter in Iranian) had satisfactory methodological credentials. The estimate was 0.96%, with 95% certainty of the true value being between 0.65 and 1.44. The estimate included stuttering of acquired neurogenic origin.

### A large data set

There are data about stuttering among 3–17-year-olds that come from analysis of the extensive United States National Health Interview Surveys (N=119,367),<sup>182</sup> which is the principal source of health information about United States citizens. It includes a range of developmental disabilities: learning disability, autism, ADD/ADHD, cerebral palsy, hearing impairment, visual impairment, intellectual disability, and seizures. Something about this study makes it more believable than other reports of stuttering prevalence. Parents were visited for an interview and were asked if “a doctor or health professional” (p. 1035)<sup>182</sup> had ever told them that their child had one of those disabilities. This, at least to some extent, gets rid of a common problem with this type of population research: inaccurate self-identification, or inaccurate identification by others such as parents or teachers. In this data set, a “doctor or health professional” reportedly made the diagnosis.

The study<sup>182</sup> indicated a point prevalence of 1.6% for stuttering, which is much higher than the estimate of 1.2% from the cross-sectional studies of children discussed earlier. Of all the developmental disabilities in that study, stuttering was the equal ranked third most prevalent.

But still, the results from that study<sup>182</sup> might be an underestimate, regardless of any merit with its methods. One reason is that a minority of parents of young children with communication disorders seem to seek health care advice about them.<sup>194</sup> Therefore, they may not necessarily find themselves in a situation to be told by a doctor or health professional that their child has a communication disorder. Another reason is that children younger than 3 years were excluded from the data set, yet some information to be discussed shortly shows that many cases of stuttering begin earlier in life than that.

### A Japanese data set

At 3 years of age, around 95% of Japanese children receive public health checkups. A report used a data base of such children to screen and confirm stuttering.<sup>195</sup> The researchers surveyed 2,448 guardians and received 1,988 valid responses. There were 465 who children screened positive to the survey, and 44% of those were assessed for stuttering by speech-language pathologists. Some of those assessments were done by observing the children, and some were done by interviewing the parents. However, it is not clear from the report how many children were directly assessed by the speech-language pathologists. The other 46% of children who screened positive were followed up with a questionnaire or telephone interview with parents. Results indicated a prevalence of 6.5% in 3-year-olds.

## CUMULATIVE INCIDENCE OF STUTTERING

### Cumulative incidence

*Cumulative incidence*, sometimes referred to simply as incidence, is the number of new cases of a condition during a certain period. It does not include recoveries during that period. So, for example, the cumulative incidence of a condition up to 12 years of age remains the same regardless of how

many recovered from it. Sometimes cumulative incidence is discussed without specifying the period, or without reference to recoveries, which can be confusing.

### Childhood cumulative incidence

#### *A caveat*

The most rigorous way to determine childhood cumulative incidence of a condition is with prospective epidemiological methods, which study its developmental course with a cohort of children. In the case of stuttering, however, a study of natural development has methodological challenges. Any such attempt by necessity must alert parents to the first developmental signs of stuttering. Yet, as stuttering develops naturally in the community, not all parents will have such awareness; therefore, such studies have a fundamental validity problem that they are not really studying the natural developmental course of early childhood stuttering.<sup>†</sup> There have been three longitudinal studies published for stuttering, which are discussed next.

#### *The 1,000-family study*

A prospective study of a cohort of children published in 1964,<sup>196</sup> known as the *1,000-family study*, is an epidemiological landmark of the field. Children born in Newcastle-Upon-Tyne, England, during May and June of 1947, were assessed regularly for a range of health conditions. The table presents cumulative stuttering incidence at various ages. The following table incorporates information provided in another source<sup>197</sup> (p. 10) about the numbers of children in the cohort at various ages, and the data from Figure 3 (p. 32) of the original publication<sup>196</sup> about the number of children who were stuttering. As mostly occurs with longitudinal studies, the participant numbers decrease over time, which is known as participant attrition.

AGE	PARTICIPANTS	NUMBER STUTTERING	CUMULATIVE INCIDENCE
<i>Birth</i>	1,142		
<i>1 year</i>	967	0	0
<i>5 years</i>	847	30	3.5%
<i>15 years</i>	763	9	6.6%

Albeit a landmark study, and decades ahead of its time, the methods of stuttering identification used for the 1,000-family study are currently recognised by modern standards as a serious weakness that damages the credibility of its results.<sup>102,198</sup> Rather than clinicians, stuttering was identified as present or absent by “health visitors” with a nursing background, who are a feature of the British medical health system.

#### *The Bornholm studies*

A more recent large-scale longitudinal study<sup>199</sup> involved all 1,042 children born during 1990 and 1991 on the Danish island of Bornholm. The health services of this island included “a free speech and hearing evaluation” (p. 49)<sup>199</sup> by a speech-language pathologist. Parents of all the children were recruited just prior to their third birthday, and 1,021 parents agreed to participate in the study and receive the evaluation.

The study did not involve subsequent, identical longitudinal assessments to identify later cases of stuttering onset. Instead, 5 years later, when the children were 8 years old, the researcher inspected the school records of the children for indications of stuttering and interviewed “various community

<sup>†</sup> Thanks to Ross Menzies for this critical point.

people, such as nurses, social workers, and teachers, who were in position to know about the children” (p. 51).<sup>199</sup> Then, 4 years later, when the children were 12 years old, “all four clinicians who cover the island’s entire school population were interviewed by the author and were asked to examine their records for any indication of new stuttering cases” (p. 51).<sup>199</sup>

The report indicated a 3-year cumulative incidence of 5.0%; 51 of the 1,021 children were stuttering at 3 years of age. At 8 years of age two additional cases were identified, giving an 8-year cumulative incidence of 5.2%. The 12-year cumulative incidence remained unchanged at 5.2%. Of the child stutterers, 52% were boys and 48% were girls.

In the review article mentioned previously<sup>102</sup> the authors describe a subsequent Danish publication from Bornholm<sup>200</sup> that is not available in English. They indicate that the original author reported another study of

928 children, comprising 92% of the island’s newly born children during a different set of two consecutive years ... each child was individually evaluated soon after his/her 3rd birthday. The same criteria for stuttering as in the first (2000) study were employed but the procedures were more direct. Specifically, the children’s speech samples were audiotaped and evaluated by the examiner to verify the presence of stuttering and to rate its severity ... [the researchers] identified 176 children who stutter ... 101 boys and 75 girls, yielding a 17.7% [3-year cumulative] incidence. Whereas one is inclined to doubt such a high figure, we emphasize that, in our judgment, very careful procedures, surpassing those of the first Bornholm study, as well as other many previous studies, were employed, including diagnosis of active stuttering by both parents and two speech-language clinicians, or detailed parent reports of past stuttering ... the current first author had the opportunity to observe several identification sessions conducted on Bornholm and can testify to the thoroughness of the procedures. (p. 71)<sup>102</sup>

#### *The Early Language in Victoria Study (ELVS) reports*

The children in this report were part of a cohort study of child language development in Melbourne, Australia: the Early Language in Victoria Study (ELVS).<sup>201,202,203</sup> The study was a prospective community cohort design, which means that the children were recruited before stuttering onset and studied longitudinally. There were 1,911 children recruited beginning at 8 months of age, with repeated observations at each subsequent birthday. The ELVS cohort was recruited randomly during 2003 and 2004 from more than 80% of Melbourne parents who visited a maternal and child health nurse when their child was 8 months of age.

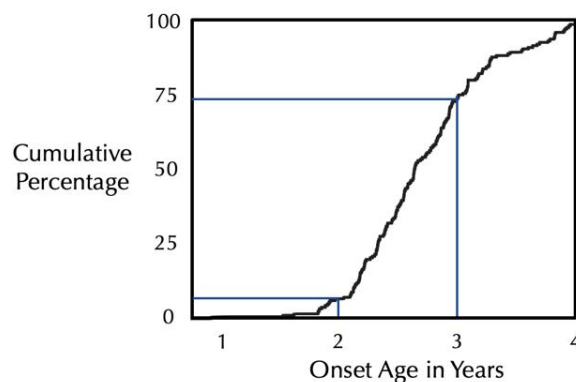
The study of stuttering within the ELVS cohort began when the children were 2 years old, and recruited 1,619 children, which was 85% of the original sample. The parents were sent a fridge magnet (pictured) describing stuttering and stating, “if you think that your child is stuttering please contact the ELVS team ... as soon as you notice it” (p. 276).<sup>204</sup> The parents were sent reminder letters about the study every 4 months for 12 months.

When a parent reported stuttering onset, a clinician visited the home, recorded a case history, and made a video recording of the child’s speech. By 3 years of age, 158 parents reported stuttering onset. The visiting clinician confirmed the presence of stuttering for 137 children and was unsure about 21 cases. A panel of clinicians reviewed video recordings for these 21 cases and agreed that their stuttering should be considered “borderline.” For the study analyses, these children were classified as nonstuttering.



The first ELVS report<sup>204</sup> showed a 3-year cumulative incidence of 8.5%: 137 of 1,619 children. This is consistent with the Japanese cohort described earlier,<sup>195</sup> which calculated a 3-year cumulative incidence of 8.9%. A subsequent report of the cohort 12 months later,<sup>205</sup> without any participant attrition, gave a 4-year cumulative incidence of 11.2%: 181 of 1,619 children. That represents one in nine children. Of the child stutterers at that time, 59% were boys and 41% were girls.

The graph\* shows the cumulative incidence of parent-reported stuttering onset by age, with specific values for 2 years, 3 years and 4 years of age. The graph conveys that the peak period for stuttering onset is between 2 and 4 years. Note that between 3 and 4 years the cumulative incidence plot is still rising but flattening, suggesting that onset rate is slowing but that more cases will appear after 4 years later in childhood.



### Conclusions about childhood cumulative incidence

Perhaps the most cautious interpretation of the prospective studies of the matter to date is that an exact figure for childhood cumulative incidence of stuttering has yet to emerge. The estimate for a 3.5% 5-year cumulative incidence from the 1,000-family study is the least believable among estimates because of the generally acknowledged limitations of that early work by modern methodological standards. It is puzzling that the two Bornholm studies, from the same research team using similar methods, would produce such discrepant 3-year cumulative incidence figures of 5.0% and 17.7%. The ELVS community cohort, with data collection prior to stuttering onset, produced a 3-year cumulative incidence of 8.5% and a 4-year cumulative incidence of 11.2%. On balance, considering that the Bornholm studies began at 3 years and may have missed children stuttering before that, the ELVS reports may be the most believable. But, regardless of what the eventual correct figure proves to be, it is clear at present that stuttering during early childhood is common.

### Lifetime cumulative incidence

The *lifetime cumulative incidence* of stuttering is the risk of being affected at some time during life, including transient periods. In a reference text<sup>101</sup> there is a review of studies where 44,129 people in total were asked whether they had ever stuttered. The authors concluded that

... it would seem that a plausible figure for the lifetime incidence of all those who at some time in their lives, either consider themselves or are considered by their parents to be PWS, is at least as high as 8% to 10% ... (p. 58)

The authors of a separate review article mentioned earlier<sup>102</sup> agree with that conclusion. In other words, considering any brief periods of stuttering during childhood, and recovery with and without treatment, there is a one in ten chance, and possibly more, that a person will experience stuttering at some time during life.

## STUTTERING ONSET

### Onset occurs during the early years of life

There is a consensus that stuttering onset occurs during the early years of life,<sup>101</sup> and this consensus comes from a review of many studies about parent recall of stuttering onset. There have been some studies of parent interview shortly after onset. A study of 10 children 2–3 years old within 2 months of

\* Adapted and reproduced with permission: Reilly, S., et al. (2013). Natural history of stuttering to 4 years of age: A prospective community-based study. *Pediatrics*, (132) 460–467. © 2013 American Academy of Pediatrics.

reported onset had a reported mean onset at 30 months.<sup>206</sup> Another study reported information about 22 child stutterers, up to their fourth birthday, who had been stuttering for up to 1 year.<sup>207</sup> The children presented with stuttering at a speech clinic. All children reportedly had begun to stutter by 36 months, with mean onset age 28 months.

These results are consistent with the ELVS report of 3-year cumulative incidence,<sup>204</sup> which reported median onset of 30 months. In that study, the lower end of the interquartile range of stuttering onset was 27 months, with the lowest onset reported at 12 months. There were 137 cases reported by parents to have occurred before 3 years of age, with 11 parents reporting stuttering before 2 years of age. The median onset age for the 4-year cumulative incidence ELVS report<sup>205</sup> was 31 months.<sup>†</sup> These results were consistent with a report of 87 children,<sup>212</sup> mean age 39 months, whose parents were interviewed before 1 year post reported onset. The mean reported onset age in that study was 33 months. A report of another cohort of 58 children by the same researchers<sup>208</sup> was a mean onset of 35 months with a range of 19–68 months.

A caveat to consider here is that those studies were not designed to detect cases of stuttering that might occur during the age range 5–11 years. Judging by the 4-year cumulative incidence ELVS graph shown previously, it seems quite possible that such onsets will occur. With the reservation about its methodology in mind, the 1,000-family study reported a rise of cumulative incidence from 3.5% at 5 years to 6.6% at 15 years. However, the first Bornholm study, which was more methodologically believable, reported 3-year cumulative incidence of 5.0%, but a 12-year cumulative incidence of only 5.2%. Regardless of what the eventual correct data will be, it is probably safe to say at present that the bulk of stuttering onset occurs during the early years of life.

### Onset can be sudden and severe

Phonological or language disorders are present when a child first starts to communicate,<sup>209</sup> but this is not so for stuttering. It appears unexpectedly after children have begun speech and language development. This can be distressing for parents,<sup>210,211</sup> particularly when stuttering onset is sudden. Reports show that half of cases become noticeable by parents within 1 week and a third during a single day.<sup>207,212</sup> The ELVS community cohort replicated these findings,<sup>204</sup> reporting that 50% of cases developed during 1–3 days and 37% during a single day. It occurs sometimes that parents report a child going to bed speaking typically and at breakfast being severely affected by stuttering. There are reports of stuttering suddenly beginning during an unremarkable day. One of many available case studies describes such sudden onset.<sup>213</sup> Stuttering is not necessarily slow to develop in terms of severity either. Severe stuttering has been reported shortly after onset, including fixed postures and superfluous behaviours.<sup>206,214,215</sup>

### Repeated movements are prominent at onset

Historically, reports have associated repeated movements with early onset. An influential 1932 account of the early stages of stuttering described them as routine.<sup>216</sup> More modern reports substantiate their prominence at onset.<sup>217,218</sup> One study<sup>206</sup> reported nearly all of 22 parents stating that their children repeated whole words and syllables at onset, and another<sup>204</sup> reported that 71% of parents recalled repeated movements at onset. But of course, if a parent reports repeated movements, it does not mean that various kinds of fixed postures and superfluous behaviours did not also occur during the stuttering moments that had repeated movements.

### Is stuttering onset predictable?

#### *Statistically*

From a statistical viewpoint, the ELVS prospective cohort study provided a few positive results.<sup>204</sup> At 3 years of age, being a boy, being a twin, having advanced vocabulary, and having a mother with advanced maternal education, were significantly associated with stuttering onset. At 4 years of age the

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<sup>†</sup> That median onset figure is not reported in the paper.

results were the same,<sup>205</sup> with two minor exceptions. Advanced vocabulary did not predict stuttering onset. However, scores for the Communication and Symbolic Behaviour Scales<sup>219</sup> were higher for children who began stuttering, suggesting more advanced communication development. So, the answer to the question is “yes,” according to the ELVS data, there are statistical predictors of stuttering onset.

The authors of the ELVS reports argued that the findings about advanced vocabulary and advanced maternal education can be explained, but the finding about twinning is puzzling. Another report from the United States National Health Interview Surveys showed that, along with many developmental disabilities, stuttering was associated with low birth weight.<sup>220</sup> However, a British study<sup>221</sup> of three birth cohorts with more than 56,000 children showed no association between stuttering and birth weight.

Research about statistical prediction of stuttering onset is rare, and there seem to be only two other pertinent reports predicting stuttering onset with reasonable participant numbers. One was a study of 87 children with early stuttering, mean age 39 months.<sup>212</sup> The researchers studied them retrospectively an average of 5.8 months after reported stuttering onset. A positive finding was reported, consistent with the ELVS cohort study, that gender was associated with stuttering onset.<sup>†</sup>

Another report<sup>222</sup> followed 96 children who were genetically at risk to begin stuttering and a control group. Twenty-six of the at-risk group began to stutter. The children who started to stutter had significantly faster articulatory rates than the children who did not; however, that difference was not significant 1 year later. Additionally, no associations were found between the linguistic skills of the children who began to stutter and those who did not. No differences were found for maternal communication style between the two groups.

### *Clinically*

Statistical significance does not necessarily mean clinical significance (as will be discussed in more detail during Lecture Five). The variables that predicted stuttering onset in the first ELVS report at 3 years were able to statistically account for only 3.7% of the cases of stuttering onset in the cohort. The predictors for the 4-year study were only able to explain 3.3% of the variance. So, the short answer to that question is “no,” according to the ELVS data, no variables were able to explain a clinically important proportion of stuttering onset.

Hopefully, future research will reverse that situation. It would be extremely useful to predict a clinically important portion of stuttering onsets. Such knowledge would have considerable public health benefits. For example, parents of high-risk children could be told of the risk and be monitored for onset by a clinician so the best early treatment could be provided at the right time. Or high-risk children could be given preventive treatments before stuttering onset. However, for now, not enough is known to allow any of that.

## **NATURAL RECOVERY FROM EARLY CHILDHOOD STUTTERING**

### **What is the natural recovery rate?**

The next issue is how many children who begin stuttering early in life will recover naturally without receiving treatment. As was the case with cumulative incidence, the most reliable way to estimate natural recovery is with prospective studies involving repeated observations of cohorts during early childhood.

### **Three essential caveats**

#### *Is natural recovery really natural?*

A complicating factor here is that there are grounds to believe that many parents do clinically useful

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<sup>†</sup> The report does not provide statistical analysis of gender data. However, they appear to be significant: *Chi Square*=10.35, *p*=.0013.



things for early childhood stuttering, independently of any clinician input. Indeed, it would be unrealistic to expect parents to do nothing when a child begins to stutter. Several reports have indicated that parents attempt to assist children with their stuttering in various ways.<sup>196,223,224,225</sup> Commonly recurring reports are that parents appear to say “stop and start again” and “slow down” to their children when they stutter. Such verbal responses may constitute the verbal response contingent stimulation of stuttering described in the previous lecture. As such, they may well be clinically useful things for parents to do.

#### *Natural recovery and treatment bias*

During extended studies of children who stutter, it is not ethical for researchers to prevent parents from seeking treatment so that they can study the natural course of stuttering. Consequently, it is important to know how much of reported natural recovery is in fact due to treatment that parents sought and received for their children during the period that researchers were studying them. So, interpretation of natural recovery reports needs to be tempered by information about how many of the children received treatment. The review that follows shows—surprisingly—that such information is usually not available.

#### *Has recovery really occurred?*

Most of the data about natural recovery are based on self-report, not the judgements of speech-language pathologists about whether recovery occurred. As such, there is some room for doubt. This was shown with a study<sup>226</sup> of 15 participants who reported natural recovery without any treatment, and 15 controls who did not stutter. Nine of the self-reported recovery group (60%) stated “that they still had a tendency to stutter” (p. 826). Independent observers watched videos of the participants. For the control group, 84% of the observers judged that the speaker “never stuttered,” but for the self-reported recovery group, only 57% of the observers made that judgement. All this raises a possibility that a portion of those who report natural recovery from stuttering may, in fact, still have mild stuttering that is not clinically significant. And perhaps most importantly, there is a tacit assumption by researchers that natural recovery refers to an absence of all behaviours associated with it. However, a study of 254 adults<sup>227</sup> made clear that those who stutter do not necessarily see it that way at all. When considering recovery, they focused on the absence of cognitive and emotional effects of stuttering, the capacity to communicate, and having control over it.

#### *The definition of recovery*

As discussed shortly, it is fundamental to clinical practice to know how many children recover from stuttering without any treatment. But perhaps the most sobering of caveats about natural recovery was a report<sup>228</sup> illustrating that the recovery rate depends on how natural recovery is defined. Table 1 in that report shows a range of natural recovery, in 23 longitudinal studies, from 6.3% to 94.0%, with a median of 66.7%. Obviously, these disparate results are of no functional use of any kind, and to gain insight into them the researchers studied 38 pre-school children on two occasions 7 years apart; first when they were stuttering at 2–5 years old, and again when they were 9–13 years old. Four criteria to determine recovery from stuttering were applied, and the resulting recovery rates, are presented in the table.

CRITERION	RECOVERY RATE
(1) <i>Less than 3.0 %SS (percentage syllables stuttered) in video speech samples</i>	94.7%
(2) <i>Two expert speech-language pathologists judge no stuttering in speech samples and parents agree that recovery occurred</i>	71.7%
(3) <i>Criterion 2 plus child self-report of recovery</i>	55.3%
(4) <i>Criterion 3 plus 0 %SS in video speech samples</i>	13.2%

In short, determining natural recovery based on stuttering count measures in speech samples is not at all valid and will grossly overestimate recovery rate. Maximum validity is obtained by asking children whether they think they have recovered. The most stringent criterion asks that of children, and also requires zero stuttering in speech samples. Probably the most clinically useful information from this table is that 7 years after early stuttering during the pre-school years, around half of children will appear to have stopped stuttering according to speech-language pathologists observing speech samples, and according to the reports of parents and the children concerned.

These reports are consistent with others. A study<sup>229</sup> of 15 pre-schoolers followed up 9 years after diagnosis reported 73% recovery based on speech measures alone, but the rate dropped to 60% when the children were asked whether they recovered. The clinical common sense to be obtained from all this was highlighted in a study of 16 children<sup>230</sup> who were studied for a mean of 19 months after onset. The parents reported that four of the children had recovered at that time, but based on clinician judgements about the presence of stuttering in speech samples, only one child was considered recovered. In other words, relying only on parent report could result in false negative decisions about the presence of stuttering during the pre-school years. As the authors concluded,

parent report of natural recovery during the pre-school years should not be taken at face value; rather, it should be subject to confirmation by a speech-language pathologist listening to at least one audio or video recording of representative everyday conversational speech of the child concerned. (p. 56)<sup>230</sup>

### Prospective reports of natural recovery

#### *The largest cohorts*

The earliest prospective report of natural recovery was from the 1,000-family study,<sup>196</sup> which indicated that 34 of the 43 child stutterers (79%) had recovered by age 16 years. There was no report of treatment history. As noted earlier, though, there are reasons to be wary about the results from this cohort.

The best-known prospective study of natural recovery is the Illinois Early Childhood Stuttering Project,<sup>231</sup> which followed 84 children for 4 years post onset and reported that 74% recovered naturally.<sup>232</sup> Although parents “were informed about availability of clinical services in the area” and that “parents decided if and when their child received treatment for stuttering” (p 1101),<sup>232</sup> the report provides no detailed information about any treatment received by the recovered children.

The Illinois group reported results from another cohort of 81 children,<sup>233</sup> of whom 58 were retained in the study for 4–5 years post onset. At that time, 39 were reported as recovered and 19 as persistent, for a recovery rate of 67%. The report contains no mention of treatment history for the children. The first Bornholm study<sup>199</sup> reported that, of the 51 child stutterers at 3 years of age, 36 (71%) had recovered at 5 years of age. Again, no information was provided about treatment history of the recovered children.

A report from the ELVS cohort of natural recovery at 7 years<sup>234</sup> presented recovery data for a portion of the children who were recruited at 2 years of age. Of 181 children confirmed to be stuttering at 4 years of age, 103 were studied at 7 years, and 67 had recovered. This provided a recovery rate of 65% at 7 years of age. Within that cohort at 7 years, 39 parents provided reports about stuttering treatment. For the children with persistent stuttering, 17% reported some kind of treatment during early life. Fewer parents of the children who recovered from stuttering—13%—reported that their child received treatment.

A large cohort report from the Twins Early Development Study<sup>117</sup> in the United Kingdom asked parents with a questionnaire at ages 2, 3, 4, and 7 years whether their children were stuttering. With the caveat that parent report has limitations, 1,085 children were reported to be stuttering on at least two of those assessments, 950 (88%) appearing to have recovered naturally. Again, no data about treatment history were reported.

A study of 38 Icelandic children<sup>235</sup> followed them from the pre-school years (mean age 4 years 3

months) through to early adulthood (mean age 18 years 6 months). This report had the most comprehensive assessments of stuttering status yet reported, which involved parent and self-report, and %SS measures. Based on parent and self-report, and a recovery criterion of less than or equal to 3.0 %SS, the recovery rate was 66%, which is two thirds. This is a much lower figure than for previous reports. Even so, there are grounds to believe this figure to be an overestimate of natural recovery because the authors reported that 81% of participants recalled having receiving treatment, which may have accounted for some portion of the recoveries. Additionally, the criterion of less than or equal to 3.0 %SS for recovery seems to be a liberal one.

#### *Smaller cohorts*

A report of 23 children with early stuttering<sup>236</sup> indicated that 16 of them (70%) had recovered 6 years later. Again, though, no details were provided about treatment history. A study of 22 children<sup>237</sup> with mean age 4.2 years reported that 15 (68%) recovered during a 2-year period. After the first year “parents had the option of continuing only observation and testing or having their child receive treatment” (p. 112) but no information is provided about how many recovered children received treatment.

The prospective study of 15 children with early stuttering mentioned earlier<sup>229</sup> followed them up from diagnosis to 9 years later and reported that 11 of them (73%) had recovered. There were two innovative parts to this study. First, complete treatment reports were presented, and all but one of the children had received treatment since their follow-up. This highlighted the impossibility of determining with this type of study what is recovery from natural factors and what is recovery from treatment. Second, the authors asked the children whether they thought they had recovered, and when they did so it appeared that only six of them (60%) might have recovered.

Another report<sup>238</sup> is worth noting, although it was not a prospective study, but a retrospective report of children diagnosed earlier as stuttering. Of the 15 cases aged 2–5 years, 12 (80%) had recovered by 7 years, although no treatment history was reported.

The only discordant prospective finding about natural recovery<sup>239</sup> involved a follow-up of 22 children with early stuttering who were diagnosed in a speech clinic and whose parents declined treatment. Eight of them were younger than 6 years at assessment and all were found to be stuttering 6–8 years later. Considering the issue discussed earlier about the confound of treatment in natural recovery studies, it is intriguing that this is the only report with information about treatment history; none of the eight children received treatment and none recovered.

#### *Conclusions*

Taken together, these findings suggest that, after onset during the early years of life, around two-thirds to three-quarters of children will recover at some later time. However, exact figures about how many children recover, and when they recover, have yet to be reliably determined. Overall, the findings about natural recovery are confounded by unknown treatment histories of recovered children. It is also problematic that nothing is known about whether parents of the children in recovery studies made any therapeutic responses themselves to their children’s stuttering.

### **Prospective reports of natural recovery soon after onset**

#### *Why is this important?*

Lectures Six and Seven show that the merits of early treatment for early childhood stuttering have become apparent during recent decades. Hence, it is now obvious that effective treatment needs to occur at some time shortly after onset. Clinicians cannot wait for many years in the hope that natural recovery will eventually occur. That being the case, information about the rate of recovery during the early years of life is essential to consider during clinical decision making.

#### *Four data sources*

Data from the Illinois Project<sup>232</sup> provide information about this matter. The mean age of the 84 children in this study was 40 months at recruitment. Table 4 (p. 1105) of the report shows that five children

(8.1%) had recovered at 12–17 months after recruitment.

The ELVS report of 4-year olds<sup>240</sup> indicated that nine of the 142 children (6.3%) recovered within 1 year of onset. However, that result was from a community cohort. It is possible that children from that population who would be taken to clinics would have a higher recovery rate during the first year post onset.

A randomised controlled clinical trial of early childhood stuttering treatment<sup>241</sup> also provided some indication of what the recovery rate shortly after onset might be. The trial had a control group of 25 children who received no treatment for 9 months. Three of these children (12%) appeared to recover during this period. A limitation of this data set is that it was not designed as a prospective epidemiological study of early stuttering. However, its strength is that it was a clinical group of children.

Sixteen children with a mean age of 36 months who presented at a clinic<sup>230</sup> were studied for a mean of 19 months. During that period, they received no treatment. Only one of the children (6.3%) was confirmed by parents and the speech-language pathologist to have recovered.

### *Conclusions*

Based on these studies, it seems reasonable to conclude that the natural recovery rate 9–18 months post onset is no more than 10%. In other words, it seems that there is some chance of natural recovery within 1 year post onset, but it is a quite a small chance. Those estimates are based on community and clinical samples.

## **IS NATURAL RECOVERY CLINICALLY PREDICTABLE?**

### **Reported family history**

A review<sup>101</sup> of 21 reports dating from 1937 to 2005 (Table 3-5, p. 95) shows that 88% of families of children with “persistent” stuttering had a reported family history. However, 63% of families with “recovered” children had a reported family history. That might be interpreted to mean that a family history of stuttering can predict to some (unknown) extent whether a child will recover. A study of 1,043 relatives of 66 child stutterers<sup>242</sup> reported a genetic trend for persistence and recovery from stuttering. The large cohort Twins Early Development Study<sup>117</sup> mentioned earlier replicated that finding.

### **The Early Language in Victoria Study (ELVS) cohort**

The ELVS report of 4-year-olds<sup>205</sup> explored a range of putative predictors that might explain natural recovery within the first year post onset. The most prominent finding was that boys had a greater chance of recovery with an odds ratio of 1.5 (95% *CI*=1.1–2.1, *p*=.02). That means that boys had an estimated 1.5 greater odds of recovering than girls, and the plausible range for the true odds ratio value, with 95% certainty, was somewhere between 1.1 and 2.1.<sup>†</sup> None of the girls recovered within the first year of onset, and 10% of the boys did. The intriguing ELVS finding associating stuttering onset with twinning,<sup>204</sup> recurred, with a reported odds ratio of 3.3 for twins recovering (95% *CI*=1.4–7.4, *p*=.005). That finding is consistent with the connection of a genetic link to natural recovery. The report also linked maternal education to recovery, including greater odds of recovering with a mother having a degree or other postgraduate qualification. The odds ratio was 1.8 (95% *CI*=1.1–2.9, *p*=.004). Four of the nine children who recovered in this report received professional help, four from a speech-language pathologist.

The ELVS report of natural recovery by 7 years of age<sup>234</sup> included a range of predictors: gender, family history of stuttering, language skills, temperament, child quality of life, and nonverbal cognition. Children who recovered were significantly more likely to have strong language skills than children who did not. Girls with better language skills at 2 years had better odds for recovery than girls who did

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<sup>†</sup> See Lecture Five for more details about odds ratios.

not. The odds ratio was 7.1, with a wide 95% confidence interval of 1.3–37.9. That means, in short, that the result should be considered with caution because of that wide confidence interval, and because a low odds ratio of 1.3 would not be particularly important clinically. The effect for language skills was not found for boys. No other predictors were found. Of the children who recovered, 13% received some kind of treatment for stuttering, and 17% of the persistent group received treatment.

### The Purdue Stuttering Project cohort

#### *Developmental trend of language as a predictor of natural recovery*

A report from the Purdue Stuttering Project<sup>243</sup> involved 50 child stutterers with a mean age of 57 months at the start of the study. Steep growth of syntactic development during yearly clinic visits over 3 years was reported to predict natural recovery at the end of that period. In this report, and with other reports for this cohort, judgments of recovered or persistent stuttering were based on a combination of speech-language pathologist and parent judgment, and speech measures (“Stuttering-Like Disfluencies,” to be discussed during Lecture Four).

There was an odds ratio of 11.1 (95% CI=1.9–65.4,  $p<.01$ ) in favour of children with steep syntactic development. There are two reservations about interpreting this study, apart from the usual reservations about such a wide confidence interval for the odds ratio. The first is that the children had been stuttering for some years at the time the study began. The second is that the report does not indicate how many of the recovered children received treatment. Regardless, a contribution of this report was the idea that developmental language trends, rather than static measures at one time, may be involved with predicting natural recovery from early childhood stuttering.

These results are consistent with a report from the same cohort<sup>244</sup> for 65-month old children, 19 of whom recovered and 13 persisted with stuttering. While the children watched video cartoons, EEG data were collected for an event related potential (N400) associated with lexical processing of visual material. Analysis of variance generated evidence that the children who persisted with stuttering had less advanced development of semantic processing. Eight of 19 (42%) of the recovered group had received treatment.

#### *Phonology as a predictor of natural recovery*

A smaller study from the Purdue Stuttering Project,<sup>245</sup> reported predictors of natural recovery for 40 children with early stuttering and 25 controls. The children were followed for a mean of 38 months until a median age of 7 years 11 months. Regression analyses showed two statistically significant phonological predictors of recovery: consonant production and nonword repetition abilities. Odds ratios were not reported. No language measure was a successful predictor. The authors reported that 27 of the stuttering group (68%) had received treatment at the time of their first assessment. They reported that they were unable to statistically adjust for this potential confound.

#### *Algorithm as a predictor of natural recovery*

With the same cohort, researchers<sup>246</sup> confirmed that the Stuttering-Like Disfluency taxonomy was unable to add anything to the predictive value of existing measures of whether 3- and 4-year-olds will recover. Hence, they reported a study of 4- and 5-year-old stuttering children (N=47) to determine any predictors of whether they had recovered when they were 6–9 years old. No information was provided about whether the children received treatment during the period of study. They used a complicated algorithm<sup>247</sup> based on Stuttering-Like Disfluencies to determine whether recovery could be predicted in the 4- and 5-year-olds: Weighted Stuttering-Like Disfluencies (to be discussed during Lecture Four). The algorithm is derived from coded transcripts of language samples.

The researchers reported detailed sensitivity and specificity (see *Screening for early stuttering* earlier in this lecture) results for the Weighted Stuttering-Like Disfluency score. The sensitivity of the index for the 29 children who recovered—the proportion of correct predictions—was 83.3% at a cut-off score of 4.2 (Table 2). Regression analysis showed an increased odds of 1.2 for stuttering persistence for each 1-unit increase of Weighted Stuttering-Like Disfluency scores. At the cut-off Weighted Stuttering-Like Disfluency Score of 4.2, three of the 18 predictions (17%) were false negative. In other words,

17% of children predicted to recover in fact did not.

#### *Statistical modelling of natural recovery*

Another report from Purdue Stuttering Project cohort<sup>248</sup> involved 52 child stutterers, studied from a mean age of 4 years 6 months for a mean period of 3.2 years. They had been stuttering for a mean of 1.6 years prior to the study. Thirty-one of the children recovered during the period of study. For this report, there was no indication of whether the children received treatment for their stuttering. The report involved detailed statistical modelling of recovery and persistence using several variables: reported age of stuttering onset and stuttering duration, family history of stuttering, family history of recovery from stuttering, several speech and language assessments, and the Weighted Stuttering-Like Disfluencies algorithm.

Results showed that gender, age, age at reported onset, and time since reported onset had no predictive value for persistent stuttering. Successful statistical modelling of persistence was established with four variables: reported family history of stuttering, Weighted Stuttering-Like Disfluencies, the Consonant and Phonological Process Inventory subtests of the Bankson-Bernthal Test of Phonology,<sup>249</sup> and the Nonword Repetition Test.<sup>250</sup> A multivariable model involving all variables was found to have better predictive value than individual variables. For that model, prediction accuracy in the range of 80–100% was reported. The combined error rate—false positives plus false negatives—was around 10% for the model. Removing either one of the phonology assessments (Bankson-Bernthal Test of Phonology or Nonword Repetition Test) resulted in higher error rates.

#### **The Illinois cohort**

The two prospective Illinois cohorts have contributed preliminary suggestions about the predictability of natural recovery. The first cohort<sup>251</sup> implicated language and phonological skill, genetics, and certain types of stuttering moments as predictors of natural recovery or persistence. However, these were flagged only as “promising predictors” (p. 51). The second Illinois cohort<sup>208</sup> of 81 children provided similar suggestions, adding increased variability of jaw displacement and negative temperament as predictors of early stuttering persistence. However, those results were presented with the qualification that “results were not definitive” (p. 12).

Another preliminary finding emerged from the first Illinois cohort about a connection between natural recovery and breastfeeding.<sup>252</sup> Forty-seven mothers were studied retrospectively, 30 of whose children recovered naturally and 17 of whose children did not. Data showed a statistically significant effect for boys, with an odds ratio of 0.17, indicating that 1 year of breastfeeding was associated with around one-sixth the odds of persisting with stuttering. However, the report provided no estimate of the confidence interval for the reported odds ratio, making the finding difficult to interpret. Regardless, this finding conceivably could be explained in terms of fatty acid nutrition and neural tissue development. However, the authors were suitably cautious about the preliminary nature of the finding.

#### **Two small cohorts**

The small-cohort study mentioned earlier<sup>236</sup> for 23 children with early stuttering generated data about predictors of recovery 6 years later.<sup>253</sup> For the 16 (70%) who recovered, analysis of variance was used to provide evidence of an association between lower articulation rate and simpler maternal language and natural recovery 6 years later. No details were provided about treatment history.

Another small-cohort study<sup>254</sup> involved children who were 3–4 years old at assessment: 30 who were diagnosed to be stuttering and 20 who were diagnosed to be not stuttering. The diagnoses were based on “stuttering-like disfluencies” (see Lecture Four) during a 300-syllable narrative by the children in the laboratory, and “a parental concern for stuttering” (p. 935). The children were studied repeatedly over a 2–2.5-year period until they were around 6 years old. The experimenters used complex acoustic analyses—envelope spectral analysis and empirical mode decomposition—to distinguish the children who recovered from stuttering from the children who persisted with stuttering. An exploratory study of seven acoustic measures found that two of them were able to predict the recovered children with a success rate of 77%. The experimenters speculated that this method may be of future

theoretical and clinical use.

### Two reviews of natural recovery

A systematic review of the topic<sup>255</sup> incorporated 35 studies with methodologies thought to be acceptable. A fundamental requirement for inclusion was that studies needed to include child stutterers and control children. Four variables were identified as replicated predictors of recovery from early childhood stuttering:

phonological abilities, articulatory rate, change in the pattern of disfluencies and trend in stuttering severity over one-year post-onset (p. 359)<sup>255</sup>

The authors concluded that it is too soon to draw any conclusions from this body of research because of inconsistencies in the methods employed. They concluded that “there is a need for systematic and replicated testing of the factors identified before initiating their use for clinical purposes” (p. 359). Regarding an abrupt decline of stuttering severity one year after onset as a predictor, the authors caution that:

... a significant pitfall of relying on trend in stuttering severity as a predictor is that the factor needs a waiting period of one year to predict the future course of the disorder. In the case of early recovery in stuttering, the recovery period will be within 18 months post-onset. ... a year wait period for initiating treatment for stuttering can considerably reduce the outcome of the treatment program. (p. 368)<sup>255</sup>

Another review of the topic,<sup>256</sup> published in the same year as the previous one, included children with early stuttering who were younger than 6 years and who were followed-up for at least 2 years. From 11 cohorts, 41 studies met eligibility and methodological criteria. Results were that some variables significantly distinguished children who did recover from children who did not. Children who did not recover were

(a) were more likely to be male; (b) begin stuttering at a later age; (c) have known family histories of stuttering ... (d) produce higher stuttering frequencies; and (e) perform lower on measures of speech sound accuracy, expressive language, and receptive language. (p. 3005)

The authors noted that effect sizes detected were modest, with the largest being a risk ratio of 1.9 for reported family history. This suggests that children with a family history have nearly twice the risk of persisting than children without a family history. The next largest effect size was gender, with boys having 1.5 times the risk of persisting than girls. The next largest effect size was for reported age of onset, with the persistent group having a mean of 40 months and the recovered group having a mean of 34 months.

In contrast to the previous report,<sup>255</sup> the authors stated that their findings were suitable for application to clinical practice, although they cautioned that the findings cannot be applied to individual clinical children. They argued that the results indicate the need for “comprehensive speech-language evaluations when working with young children who stutter” (p. 3009), stating that such evaluations

... may provide some guidance that is useful to clinicians as they evaluate whether a child is presenting with characteristics associated with higher risk for persistence until empirically supported cutoff scores are available. (p. 3009)<sup>256</sup>

### Clinical conclusions about the clinical predictability of natural recovery

The short answer to the question of whether natural recovery is predictable is similar to the answer to the previous question of whether stuttering onset is predictable. In a statistical sense the answer clearly is “yes.” But, as with stuttering onset, the clinical applicability of that statistical information is concurrently an important matter, and not a straightforward one. Some considerations about it are now presented.

*Natural recovery prediction has error rates*

In one sense, efforts to predict natural recovery are screening procedures, which were discussed earlier in this lecture. As such, they are associated with error rates. They have a true positive rate (sensitivity) and a true negative rate (specificity), along with false positive and false negative rates. As noted earlier in this lecture in the context of screening, a potentially harmful outcome is false negative. In the case of predicting natural recovery, this is forming a view that a child will recover naturally from early stuttering when the child in fact does not. Publications from the Purdue cohort have noted the clinical seriousness of such an error:<sup>246,248</sup> “In the case of early treatment, failing to identify a TP [true positive] may have profound, lifelong ramifications” (p. 2562).<sup>246</sup> This is because of the superior efficacy of early childhood stuttering treatment compared to treatment for persistent stuttering, as noted earlier in this lecture. A delay of early treatment based on a mistaken judgment about natural recovery could be a serious clinical error.

Regrettably then, there seems to be only one publication providing an estimate of false negatives for predicting recovery: 17%. This was for the single predictor of Weighted Stuttering-Like Disfluencies.<sup>246</sup> The most comprehensive statistical modelling of natural recovery to date,<sup>248</sup> from the Purdue cohort, does not supply information about false negatives, only “total number of errors (false positives plus false negatives)” (p. 2929).

*How should predictors be used clinically?*

Because of the inherent error rates in methods to predict natural recovery, researchers have acknowledged that nothing can be said with certainty about whether an individual child will recover naturally or not. That said, the question remains about what clinical use these clinical predictors might be. The authors of the Purdue reports, while acknowledging the clinical time required to measure children’s phonological skills and a Weighted Stuttering-Like Disfluencies score, recommend that those assessments, along with determination of family history, be routinely included in clinical assessment of pre-schoolers who stutter. That view was endorsed with a systematic review.<sup>256</sup>

However, as yet, there is no clinical guideline to emerge from this research that makes clear, when such effort is made, what should be done with the results of that clinical assessment. The Purdue researchers have offered only suggestions that they may be useful for prioritising treatment services<sup>246</sup> and deciding when to monitor a child for recovery rather than providing treatment.<sup>248</sup> The lack of clarity in this matter is arguably confounded by a statement in the latter report obviating the value of recovery prediction in any clinical case of early stuttering involving parent or child concern. (Presumably, that would be the case when most children are brought to a clinic.):

... regardless of their risk profile, if a child (or their parent) is expressing concern, anxiety, or negative feelings and attitudes toward their communication abilities, that child (and family) would clearly benefit from treatment ... (p. 2922)<sup>248</sup>

*Natural recovery prediction and the timing of early treatment*

As noted in a systematic review,<sup>255</sup> there is a tension between the need for early childhood stuttering treatment and the clinical use of existing reports about predicting natural recovery. This is because reports have studied children who have been stuttering for several years, and during that period the tractability of early childhood stuttering may decrease. In other words, waiting for some years in the hope that natural recovery will occur is potentially associated with a clinical penalty if natural recovery does not occur. Added to that issue is that a clinically significant period elapses in many cohort studies before the first observation; 1.6 years in one report from the Purdue cohort.<sup>248</sup>

Complicating this issue is that the ELVS cohort, which began when the cohort was 2 years old—much earlier than any other cohort—established recovery predictors that were different to other cohorts. For example, during that early stage of stuttering development, the ELVS cohort reported that family history was not predictive of recovery at all, and that gender was involved. This is the opposite of findings from the Purdue cohort and could be explained by the younger ELVS cohort.

Finally, it is difficult to apply existing research about predictors of natural recovery to early treatment



for the simple reason that the effects of treatment and natural recovery cannot be disentangled in that literature. For example, in the ELVS cohort, parents reported that 15% of the children received some form of treatment, and in the Purdue cohort around two-thirds of children reportedly received treatment.

Another complicating factor affecting clinical judgements about natural recovery and when to begin stuttering treatment is that stuttering is associated with a range of mental health issues, and those problems begin during childhood. The impact of that on when to begin stuttering treatment is an onerous consideration that will be taken up during Lecture Eleven.

#### *Validating prognostic models<sup>†</sup>*

It is a justifiable viewpoint, expressed in one systematic review,<sup>255</sup> that it is too soon to apply this body of research to clinical practice. It is accepted practice in health care statistics that a prognostic model needs to be validated, in the sense of being shown useful for clients other than those from whom the data were derived. A seminal paper on the topic<sup>257</sup> outlines several reasons why this is necessary. First, mathematical prognostic models are likely to provide overly optimistic estimates of how they will apply to the real world. For example, the model developed from the Purdue cohort<sup>248</sup> relies on professional, community clinicians completing a range of formal assessments. Second, mathematical prognostic models are prone to statistical error. One such source of error is limited sample size, which certainly is a consideration in the literature about estimating natural recovery rates. Finally, the model may not apply to locations other than those in which they were developed. In the case of the Purdue model from the United States,<sup>248</sup> there is much work yet to be done to establish whether it applies to other clinical communities worldwide.

## **WAITING LIST PRIORITISATION FOR CHILDREN WHO STUTTER**

Considering how common childhood stuttering is, and its potential effects on people throughout the lifespan (see Lecture One), it is not surprising that a recent report<sup>258</sup> found that speech-language pathologists around the world prioritise treatment of childhood stuttering above all other developmental speech and language disorders. The report was a survey of 264 speech-language pathologists from 10 countries: Australia (n=182), United States (n=37), United Kingdom (n=15), Canada (n=9), New Zealand (n=6), Ireland (n=4), Scotland (n=1), South Africa (n=1), China (n=1), and The Netherlands (n=1). The speech-language pathologists worked mostly with children (78%), with 89% working with 3–5-year-olds, 83% working with 6–12-year-olds, and 43% working with 13–18-year-olds. Most of the speech-language pathologists (74%) reported a waiting list in their workplace.

Results indicated that the highest waiting list priority was given to children who stutter above children with other childhood speech and language disorders. The speech-language pathologists indicated stuttering as a priority most frequently (47%) compared to disorders of language (36%), speech (30%), and voice (17%).<sup>†</sup> Reports indicated, compared to other speech and language disorders, that children who stutter most often bypassed waiting lists and immediately received assessment and treatment services. Those reports are consistent with interviews of 18 Australian speech-language pathologists about children younger than 12 years.<sup>259</sup> Those clinicians conveyed a belief that stuttering is “more debilitating than other communication disorders with the potential for long-term consequences to be more significant for clients” (p. 5). Consequently, those clinicians associated childhood stuttering with substantive professional responsibility.

## **SUMMARY**

Early and persistent stuttering are clinically significant developmental stages. Diagnosis of stuttering is generally not a clinical challenge, with adults and parents usually being correct with their

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<sup>†</sup> Thanks to Mark Jones for guidance with this material.

<sup>†</sup> The highest priority was given to feeding difficulties, at 89%.

identifications. However, there are some conditions that potentially could be mistaken for stuttering. There is an ambiguous body of literature about whether speech and language disorders are comorbid with stuttering. Genetics is involved with stuttering, although the complete picture of how is not clear at present. Children and adults who stutter have atypical speech neural processing. However, it is not yet clear how that relates to the cause of stuttering. Stuttering is a common condition that is extremely prevalent during early childhood. A comprehensive database shows stuttering to be the equal third ranked of a range of developmental conditions. Its 4-year cumulative incidence could be as high as one in nine children. Onset occurs early during life unexpectedly, unpredictably, and often rapidly. Two-thirds to three-quarters of children will recover at some later time. The most methodologically rigorous method of determining natural recovery suggests that the true figure is around two-thirds. The probability of recovery during the first year after onset is low. Preliminary work has been done to establish workable prognostic models of natural recovery. Speech-language pathologists around the world prioritise treatment of childhood stuttering above all other developmental speech and language disorders.

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## LECTURE THREE: THE CAUSE OF STUTTERING<sup>†</sup>

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### TWO REASONS CAUSALITY IS CLINICALLY IMPORTANT

#### Explaining cause to clients and parents

A fundamental way to cope with having a health issue is to understand its cause. So, an obvious way causal explanations of stuttering influence daily clinical practice is for clinicians use them to explain what stuttering is and why clients or their children are affected by it. This is particularly important with early childhood stuttering. As discussed during Lecture Two, stuttering onset during early childhood is unpredictable, and its onset can be sudden and severe. This can be distressing for parents, so an explanation of why this happens is fundamental to clinical practice.



#### Treatment credibility and expectancy

Ideally, there will be a transparent link between what clinicians explain about the nature and cause of stuttering and how they propose to treat it. A treatment that makes sense this way is likely to be more credible to clients and parents than one that makes no theoretical sense. The notion of treatment credibility is “how believable, convincing, and logical a given treatment is” (p. 27).<sup>1</sup> A related notion outlined in that paper is treatment expectancy, which refers to what clients believe can be achieved with a treatment.

There is evidence that constructs of treatment credibility and expectancy are related to parent compliance with psychological treatment for children.<sup>1</sup> This issue has been found to be pertinent for one of the childhood treatments discussed later in these lectures (the Lidcombe Program):<sup>2,3</sup> “I didn’t think that [*the treatment*] was really going to make such a difference and it did” (p. 76).<sup>3</sup>

### STUTTERING CAUSALITY IS NOT A SIMPLE MATTER

At first, it might seem that the notion of what causes something to happen is a simple matter. But this is not always the case. Packman and Attanasio’s classic reference text, *Theoretical Issues in Stuttering*,<sup>4</sup> contains a discussion about what causes a fire (Chapter 1, pp. 10–11). This gives some insight into the potential complexity of studying causality. It is clear from Lectures One and Two that stuttering is a complex matter. Considering that, and considering also that verbal communication itself is a particularly complex process,<sup>5</sup> stuttering causality is a challenging prospect to consider.

In the Packman and Attanasio text the first two chapters provide a brief introduction to the philosophy of science that deals with causality. Those chapters cover concepts such as *necessary and sufficient* conditions for something to occur, fallacies of causal reasoning, the philosopher Thomas Kuhn and scientific revolutions, pseudoscience, and paradigms. Packman and Attanasio extend Gerald Siegel’s observations<sup>6</sup> that the complexity of stuttering involves domains such as the perspective of those who seek to understand causality, the level of understanding required, and why a causal explanation might be sought.

This is not to say that causality of health issues is never simple. Packman and Attanasio mention single gene anomalies that cause conditions such as cystic fibrosis or sickle cell anaemia. All those who have

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<sup>†</sup> Thanks to Ann Packman for guidance with this material. This lecture draws extensively from Packman, A., & Attanasio, J. S. (2017). *Theoretical issues in stuttering* (2nd ed.). London, UK: Routledge.

the genetic anomaly have the condition and nobody has the condition without the genetic anomaly. Or to say it another way, a single gene is *necessary and sufficient* to have the condition.

The causality of stuttering, however, is not so simple. The concepts of *necessary* and *sufficient* when considering stuttering causality need to be considered. If something is *necessary* for stuttering, all those who stutter must have it. If something is *sufficient* for stuttering, those who stutter may have it, but won't necessarily have it. Unlike cystic fibrosis and sickle cell anaemia, there is no one factor that is *necessary and sufficient* for stuttering. Stuttering is multifactorial, and this aspect of it is resumed later in this lecture.

## THE HUMAN SCIENCES: THEORIES, HYPOTHESES, AND MODELS

In their classic reference text,<sup>4</sup> Packman and Attanasio outline a distinction between the physical and the human sciences, pointing out that the study of stuttering is in the latter category. In contrast to the physical domain, human behaviour requires a different research approach because of the intrinsic variability of humans. As stated by Packman and Attanasio, when researching about stuttering causality,

... the experiment needs to be performed with many human subjects to determine if the finding occurs in a sufficient number of subjects to be considered a meaningful effect. (p. 22)<sup>4</sup>

Packman and Attanasio refer to theories, models, and hypotheses as ways to present causal propositions used in the search for understanding. They state that, in such a pursuit, the terms "theories," "hypotheses" and "models" are commonly used interchangeably, even though there is a distinction between them in the strictest sense. Theories typically have strong evidence to support them; hypotheses are more tentative propositions than are theories. A model is "a physical and/or mathematical and/or conceptual representation of a system of ideas, events or processes."<sup>7</sup> Or, stated more simply, models explain how things work, and are commonly presented using boxes, circles, lines, and arrows. Packman and Attanasio refer to several categories of models. Some involve a linear cause-effect relationship, and some are transactional, with a bidirectional link between cause and effect.

## AN HISTORIC AND CLINICALLY INFLUENTIAL CAUSAL EXPLANATION

### The Diagnosogenic Theory

There are many early causal explanations of stuttering that are now of historical rather than scientific interest, and overviews are given in various reference texts.<sup>4,8,9</sup> Examples include pyknolepsy theory (a type of childhood epilepsy), perseverative theory, approach-avoidance theory, the Orton-Travis Theory, two-factor theory, primary and secondary stuttering theory, and psychoanalytic theory. Wendell Johnson's Diagnosogenic Theory is regarded as one of these causal explanations now of historical interest.<sup>4,10</sup> However, there is much about its influence on clinical practice that is instructive. One of Johnson's famous students, Oliver Bloodstein, gave an engaging account of the origins of this theory from the field of general semantics.<sup>11</sup>

### The rise

The fundamental premise of the Diagnosogenic Theory was the paradoxical and circular idea that stuttering is caused by its diagnosis. In short, parents caused the development of stuttering by falsely believing that their children had begun to stutter when, in fact, they had typical disfluency. According to the theory, it was when parents became anxious about typical disfluencies and tried to make their children stop doing them, that stuttering subsequently developed. Johnson famously expressed his theory by stating that stuttering begins not in the mouth of the child but in the ear of the parent. The theory was influenced by some earlier ideas of Bluemel,<sup>12</sup> and was formally proposed in 1942,<sup>13</sup> based on 46 child stutterers and 46 controls who were 2–9 years old: "stuttering in its serious forms develops after the diagnosis rather than before and is a consequence of the diagnosis" (p. 257).

Part of the extensive influence of this theory throughout the Western World and beyond can be linked to Johnson's "open letter to the mother of a stuttering child," which was first published in a parenting magazine and later in a prominent journal of the American Speech-Language-Hearing Association.<sup>14</sup> There, the advice offered to parents was:

Do absolutely nothing at any time, by word or deed or posture or facial expression, that would serve to call Fred's attention to the *interruptions* in his speech. Above all, do nothing that would make him regard them as abnormal or disgraceful. (p. 7)

Do not label Fred a 'stutterer.' If you do, you will have an almost irresistible tendency to treat him as if he were as defective and unfortunate as the label implies. (p. 7–8)<sup>14</sup>

The theory and the clinical advice that followed from the Diagnosogenic Theory attained widespread acceptance, and for decades no clinician, or nearly anyone else it seems, would ever think of directly treating early childhood stuttering by calling attention to it. The situation in the 1970s is portrayed here:

one of us presented a workshop on speech and language disorders to a group of early childhood teachers ... Stuttering was included and the presenter used the word *stuttering* when the topic was introduced ... the teacher said that they had been taught that using the "label" *stuttering* would cause a child to become a stutterer ... they had also been taught that these children were experiencing "disfluencies", that they were not actually stuttering, and that the problem would worsen and they would become stutterers once they were labelled and treated as such. (p. 49)<sup>4</sup>

## The fall

The fate of most causal explanations of something is for them to be wrong: they can't all be right.<sup>15</sup> Part of scientific development is the eventual realisation that a causal explanation is wrong, and this is what happened with the Diagnosogenic Theory. The first public proclamation that the Diagnosogenic Theory was wrong was published in 1983.<sup>16</sup>

Emerging research evidence during the 1970s strongly suggested that it was wrong. A prime example can be seen in the reports of verbal contingent stimulation of stuttering with early childhood stuttering, described during Lecture One. The most famous of those was a 1972 publication<sup>17</sup> showing that two young children reduced stuttering when attention was called to it, and a similar finding to the same effect was published some years later.<sup>18</sup> If the Diagnosogenic Theory was correct, then calling attention to the children's stuttering would have worsened it, not improved it. During the early 1980s the longitudinal Illinois Early Childhood Stuttering Project<sup>19</sup> (see Lecture Two) began to produce data that challenged the theory: the speech of stuttering pre-schoolers and their typically developing peers was completely different.

Rather dramatically, the theory predicted that if a culture could be found with no word for stuttering or concept of what it was, then there would be no stuttering in that culture. Johnson published a report in 1944<sup>20</sup> stating that the Bannock and Shoshone Indians of Idaho in the United States had neither any word for nor concept of stuttering, and consequently none of them stuttered. Correspondence came to light in 1981<sup>21</sup> that Johnson was informed at the time that he was wrong, and that the tribes in question had 18 ways of referring to stuttering. When prefacing the fourth edition of his landmark text in 1987,<sup>22</sup> Oliver Bloodstein announced that the period since the previous edition had seen some "notable surprises," one being "the discovery that American Indians of the Great Plains do stutter and probably did stutter a generation ago, when they were reported not to."

There was much controversy connected with the discrediting of the Diagnosogenic Theory. As mentioned during Lecture Two, Johnson conducted an unethical experiment during the 1930s to test the theory. It was not published,<sup>23</sup> but decades later was found to not support the theory.<sup>24</sup> The experiment later resulted in legal proceedings.

## A lasting influence

*Johnson's "open letter to the mother of a stuttering child" that was mentioned earlier<sup>14</sup> offered the following advice to parents:*

Give Fred a chance to speak without interrupting him unduly with your own remarks ... See that his brothers and sisters are not always 'bossing' him or not always talking when he wants to talk ... In reading or speaking to Fred, be calm and unhurried, enunciate clearly and avoid a high, tense voice. ... When you take him to strange places or ask him to do something that is new to him, prepare him for it by explaining ahead of time. ... in general, try to avoid situations that are unduly frustrating, exciting, bewildering, tiring, humiliating, or frightening ...<sup>14</sup> (p. 8)

Today, more than six decades later, these recommendations figure in modern sources of advice to parents from professional associations and health services:

Having a short (5 minutes) one-to-one time with your child on a regular basis, when you are both calm and not in a rush and you are not likely to be interrupted ...

Thinking about your child's general well-being, his sleeping and eating habits, his health and his pace of life ... Looking at your family's conversations - are you letting each other finish what you want to say? Is anybody hogging all the talking time? Do you interrupt each other when trying to speak? ... Building your child's confidence by focusing on what he is doing well and praising him for this ... What kind of language are people using when they talk to him?<sup>25</sup>

Reduce your own rate of speech. It's common to hear parents and other adults tell children who stutter to "slow down." ... Reduce the number of questions you ask in succession. ... it's easy to slip into the role of "teacher" and inadvertently pepper them with questions to test their knowledge. ... Rather than constant questioning, try making statements beginning with "I think" or "I wonder" as a conversational prompt. ... Follow the child's lead in play. Following your child's lead helps reduce the amount of verbal instructions and questioning during play. ... Decrease length and complexity of language.<sup>26</sup>

As discussed shortly, these clinical notions are current today in clinical applications of some models of stuttering causality.

## A sobering reflection

The Diagnosogenic Theory provides a telling illustration not only of how a causal explanation can influence clinical practices, but how that influence can go awry. A report shows that, decades later, when the theory is known to be wrong and scientifically discredited, some clinicians still believe it to be true. A 2014 study of 37 speech-language pathologists and 70 speech-language pathology students<sup>27</sup> reported that "more than half of the participants indicated that parents are the primary etiological factor in stuttering and the word 'stuttering' should be avoided" (p. 778). A 2020 report indicated that 141 speech-language pathologists in the United States were reluctant to use the term "stuttering" or to diagnose stuttering with pre-schoolers.<sup>28</sup> A report of a 2019 European conference about stuttering treatment<sup>29</sup> is also telling. Delegates from 29 countries at that conference recognised the need for early treatment, and acknowledged the compelling evidence base showing that early treatment is efficacious (as discussed during Lecture Seven). Yet, there was a view that early childhood stuttering treatment would cause children to become anxious, even in the face of research evidence that this does not occur. The conference convenors ventured that the only explanation for this view could be the lingering influence of the Diagnosogenic Theory. Perhaps, then, it is not surprising that parent guilt seems to recur during accounts of early childhood stuttering<sup>30,31</sup> and (as discussed during Lecture Seven) during its treatment.<sup>32,33</sup>

## TESTING CAUSAL EXPLANATIONS OF STUTTERING

Packman and Attanasio<sup>4</sup> discuss four ways to evaluate a causal explanation: testability, explanatory power, parsimony, and heuristic value. The first two of these are now overviewed.

### Testability

To be of any value, a causal explanation needs to be falsifiable. The prime source of information used to evaluate causal explanations, and potentially falsify them, is experimentation. To cut short a long story, experimentation involves observations in contrived circumstances that make them more powerful. For example, imagine a proposition that states that the cause of stuttering is in the larynx with vocal fold function. There has been such a proposition that attained notoriety in the past.<sup>34</sup> An experiment could explore that notion, by having those who stutter speak using their vocal folds and without using their vocal folds; during lipped speech, when there is no vocal fold function, and during standard speech, when there is vocal fold function. If the proposition is correct and stuttering is a problem with vocal cord function, there should be no stuttering during lipped speech. In fact, such experiments have been done<sup>35,36</sup> and it is obvious that stuttering can occur during lipped speech. Hence, the causal explanation that stuttering is a vocal cord problem is falsified.

The influential philosopher Carl Popper is credited with the axiom that experimentation does not prove a scientific theory to be correct: it only fails to disprove a theory. However, experimentation can provide results that may be interpreted as disproof of a theory, as shown in the case above. A theory that constantly resists active attempts to disprove it attains increasing credibility. In science, a theory that resists active disproof for a long period can become known as a law. For example, the theory of gravitation is often referred to as the law of gravitation. (But even gravity, it seems, is not immune to observational challenges.<sup>37</sup>)

### Explanatory power

The more that a causal explanation can explain about its topic, the more credible it is. Stuttering presents so many things that need to be explained, and a causal explanation of it needs to be evaluated considering how well it covers them. Packman and Attanasio<sup>4</sup> argue that, ideally, the explanation needs to be done parsimoniously: that is, simply. In the case of stuttering, arguably there are some well-known features of stuttering that need to be taken account of in causal explanation. These are outlined below.

#### *Behavioural diversity*

Stuttering is behaviourally diverse. Why does it have such a range of behavioural manifestations involving different types of repeated movements, fixed postures, and superfluous behaviours? Even more challenging for causal explanation is that everyone who stutters does so in a different observable way. They have different types and combinations of the seven stuttering behaviours described during Lecture One.

#### *The influence of spoken language*

As outlined during Lecture One, stuttering moments are not random but tend to occur more on consonants than vowels, and mostly on initial sounds of words and on initial words of utterances. Those who stutter often encounter idiosyncratic difficulties with certain sounds and words. Stuttering occurs more commonly on content than function words. And a most obvious but commonly overlooked fact for any causal explanation to accommodate is that stuttering does not occur on every syllable spoken; it presents as an intermittent occurrence involving stuttering moments. This sets it apart from the continuous presence of other speech and language disruptions. For example, lisping will occur on each occurrence of affected sounds. It is even more challenging for causal explanation to accommodate the fact that language is not even necessary for stuttering to occur; stuttering can occur experimentally on non-words, where lexical processing is not necessary.<sup>38,39</sup>

### *Onset during language development*

Obviously, early developing language is a causal factor for stuttering. However, as discussed during Lecture Two, stuttering does not start when children first begin to speak. Children speak without any problems for an early period of their language development, then stuttering begins sometime later during language development. A causal explanation of stuttering needs to cover this. In this case, developing language is *necessary* for stuttering development, but not *sufficient*; all children who stutter have some language development, but most children with language development do not stutter.

### *Epidemiology*

As outlined during Lecture Two, stuttering begins during the first years of life, but why not later in life? Why does it sometimes resolve naturally but sometimes becomes a lifelong problem? Why does it sometimes begin abruptly and sometimes gradually, and why are repeated movements often among the first signs of stuttering? A causal explanation of stuttering needs to take account of all this.

### *Conditions that reduce or eliminate stuttering*

There are diverse fluency-inducing conditions, as outlined during Lecture One. Stuttering nearly always vanishes when people sing or speak in rhythm, or when they speak under chorus reading or shadowing conditions. Stuttering decreases with verbal response contingent stimulation, and under conditions of altered auditory feedback and masking. How can such a range of diverse conditions reduce or eliminate stuttering?

### *Stuttering and wind musical instruments*

Playing wind instruments has in common with speech that it involves respiratory activity combined with tongue and lip movements. There are intriguing reports, dating from the early 1950s, that some who stutter appear to do so when playing a musical wind instrument.<sup>40,41,42,43</sup> One of those reports<sup>43</sup> provided acoustic evidence of this occurring. So, causal explanation of stuttering must deal with how it can affect a non-speech activity.

### *Stuttering and manual tasks*

Although there have been findings to the contrary, there are research findings that signs of stuttering are to be found outside the speech mechanism. All this is even more intriguing than findings about playing wind instruments and speaking nonsense words, because the experimental tasks involved seem to have nothing at all to do with speech.

Examples include delayed manual reaction times for those who stutter<sup>44,45,46</sup> and finger movement tasks.<sup>47,48,49</sup> There have also been recurring reports that those who stutter do not perform as well as controls with bimanual motor sequences. One research group has found this to occur with finger tapping, key pressing, handle turning, and even peddle pushing.<sup>50</sup> Such results have been independently replicated.<sup>51</sup> Implicit motor learning refers to learning of complex motor skills over time, and presumably independently of awareness. There is evidence that, with learning finger sequencing skills on two occasions, adults who stutter do not perform as well as controls.<sup>52</sup> However, with explicit motor learning involving awareness, with a finger-to-thumb opposition sequencing task, there is evidence that those who stutter do not differ from controls.<sup>53</sup>

Compared to controls, those who stutter have been shown to have more timing asynchrony when playing piano melodies.<sup>54</sup> There is a body of research dealing with the capacity of those who stutter to synchronise with a metronome beat. Mixed results have occurred in studies of stuttering and nonstuttering adults. That literature is reviewed in the introduction to one study that reported a finding that adult stutterers did not perform as well as controls in following a metronome beat with finger movement.<sup>55</sup> A comprehensive 5-year longitudinal study of pre-school children with a rhythmic clapping task showed no differences between 70 child stutterers and 45 controls.<sup>56</sup> This raises the prospect that any such findings with adults are an epiphenomenon involving the effects of stuttering rather than its causes.



*Stuttering severity is variable*

As outlined during Lecture One, stuttering severity is notoriously variable. It is likely to vary with differing audience sizes and types, across different everyday situations, and when talking alone. As such, it is different from many conditions where presenting features remain stable over time. Causal explanation of stuttering needs to account for this feature of it.

*Genetics*

Any causal explanation of stuttering needs to incorporate how genetics contributes to cause. As outlined during Lecture Two, genetics is obviously involved causally with stuttering, although details are not fully known at present. Lecture Two makes clear that its causal role is that it is neither *necessary* nor *sufficient* for stuttering; some who stutter appear not to have genetic involvement, and some who have genetic involvement do not stutter.

*Disrupted neural processing of spoken language*

As outlined during Lecture Two, many research findings have connected stuttering with disruptions of brain networks that support speech motor control. As discussed there, definitive research has yet to be done to establish whether such disruptions are causal; it is not yet clear whether they are *necessary* for stuttering, so that all those who stutter must have them. And as pointed out during Lecture Two, such disrupted neural processing of spoken language is not *sufficient* for stuttering because children speak without stuttering for a period during language development before stuttering begins.

Regardless, the research presented in Lecture Two carries a strong implication that disrupted neural processing of spoken language might be *necessary* for stuttering. So, arguably, it is justifiable to list “disrupted neural processing of spoken language” among established causal factors for stuttering, albeit in a tentative fashion.

That said, it bears stating that a causal relation between disrupted neural processing and stuttering may not be straightforward. To use the terminology presented by Packman and Attanasio,<sup>4</sup> a model of how atypical neural processing is involved in stuttering causality may not be unidirectional; it may be transactional, with a bidirectional link between cause and effect. Organic issues with atypical neural processing may exist prior to stuttering onset, and the speaker’s subsequent efforts to compensate for them may change them in some way. Arguably, that possibility is a reality considering recent knowledge that experience is known to drive brain development,<sup>57,58</sup> and the existence of evidence that a change to atypical neural processing occurs after behavioural stuttering treatments.<sup>59</sup>

**MULTIFACTORIAL MODELS OF STUTTERING CAUSALITY****Stuttering as multifactorial**

As noted earlier in this lecture, some health issues are well explained with one factor that is *necessary* and *sufficient* for them to occur. But there are many health issues for which this is not the case, such as hypertension, diabetes, arthritis, and depression. Stuttering is one such health issue. There must be more than one *necessary* and *sufficient* factor to explain stuttering. A little reflection shows that this must be so. As indicated in Lecture Two, there is strong evidence that genetics is involved in causality, but genetics alone is not sufficient for stuttering to occur. For example, monozygotic concordance is not 100%; when one genetically identical twin stutters, the other will not always stutter. Also, there is strong evidence that disrupted neural processing of spoken language is involved causally, but it alone is not sufficient for stuttering. This is obvious, because children speak without stuttering for a period until stuttering onset occurs at a median age of 31 months.

**The Packman & Attanasio 3-Factor Model of Stuttering Moments***Background*

Known generally as the P&A Model, this is a model in the true sense of the term, as outlined earlier; it explains how things work. It is influenced by Packman and Attanasio’s<sup>4</sup> observation that causal

explanations should be driven by the level of understanding required. Accordingly, the level of understanding sought by the P&A Model is why stuttering moments occur during speech throughout life. As such, it incorporates the logic that “all causal factors must be operating at every moment of stuttering” (p. 226).<sup>60</sup> It proposes to explain the factors that, together, are *necessary* and *sufficient* for the occurrence of individual stuttering moments. In so doing, it bypasses the notions of distal and proximal causality. As noted by Packman and Attanasio,<sup>4</sup> distal causes are far removed from the event being explained, and proximal causes are near to it, and they are separated by necessary steps. With the level of understanding intended by the P&A Model, genetics is a distal cause of stuttering and does not feature in it.

Packman and Attanasio developed the P&A Model,<sup>4,60,61</sup> advancing earlier thinking outlined in previous publications.<sup>62,63</sup> They acknowledged that it incorporates components developed from earlier work by Zimmermann and Wingate. They credit Zimmerman and colleagues with the notion that the speech motor systems of those who stutter may be unusually susceptible to variability.<sup>64,65,66</sup> They credit Wingate with the notion that prosody, of which syllabic stress is a part, is somehow disturbed with stuttering.<sup>67,68,69</sup> Wingate recognised that the effect of rhythmic stimulation involves changes to stress, and, specifically, that rhythmic speech reduces stress contrasts.

### Overview

The P&A Model draws on the empirically derived features of stuttering that are well established, as outlined earlier in this lecture. It proposes three factor types that are *necessary* and *sufficient* to cause a moment of stuttering. These are a factor that is necessary, a factor that triggers a moment of stuttering, and a factor that influences the threshold for a stuttering moment to occur. These factors are now outlined.

*Necessary Factor: Disrupted neural processing.* The model assumes a central nervous system issue that gives some children disrupted neural processing of spoken language. The first published account of the P&A Model<sup>61</sup> suggested atypical white matter connectivity as the likely neural processing issue, as other researchers had done.<sup>70</sup> White matter is involved generally in transmission of information, and grey matter with information processing, and the latter has also been associated with stuttering (see Lecture Two). This issue manifests as speech that is prone to perturbation. In the model, the neural processing disruption is a *necessary* but not *sufficient* condition for stuttering to occur. In other words, everyone who stutters must have it, but it is possible to have it and not stutter.

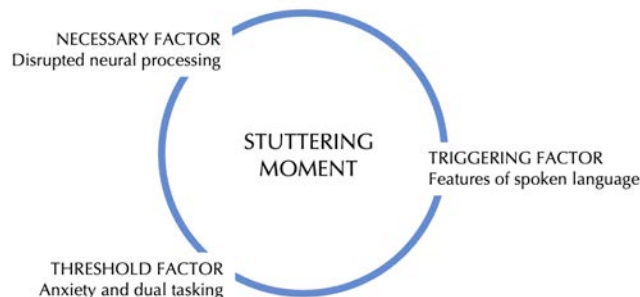
*Triggering Factor: Features of spoken language.* Moments of stuttering are triggered by certain features of spoken language. These are the variables that increase the motor task demands placed on speech production: the varying of stress (emphasis) from each syllable to the next, and linguistic complexity. This pushes those who stutter beyond what the speech system can deal with, thereby triggering stuttering moments. Hence, stuttering is not a motor speech deficit as such. This idea draws on the watershed time during early language development when children begin to produce linguistic stress contrasts. So, for example, they will say “*dad-da*,” emphasising the first syllable, instead of “*dad-da*.” That may not seem much of a difference, but it is a leap of speech production, and those early attempts to produce linguistic stress contrasts trigger stuttering moments.

*Threshold Factor: Anxiety and dual tasking.* The threshold above which moments of stuttering are triggered is modulated differently across and within individuals. The disruptive effects of speech-related anxiety are involved, which include physiological and cognitive components (see Lecture Ten). The presence of speech-related anxiety may lower the threshold for stuttering moments to occur and may be associated with more of them occurring. This draws on evidence that stuttering increases with increased audience size (see Lecture One). The model also draws on evidence that stuttering increases when dual tasking with a competing linguistic activity diverts attention away from speaking.<sup>71,72</sup> Such dual tasking would lower the threshold for triggering of a stuttering moment. An example of day-to-day dual tasking would be “attending to email, talking with colleagues, talking on the phone and taking notes, often simultaneously” (p. 943).<sup>71</sup>

The P&A Model is illustrated in the figure.\*

### *Testability*

Because the model proposes that disrupted neural processing is a necessary condition for stuttering, everyone who stutters should have that disrupted neural speech processing. The model would be falsified with future brain research that produces recurring findings of no such disrupted neural processing among those who stutter. There has been a statement of the logical prospect that the necessary condition for such disruptions means that they should be present in children, and perhaps even prior to stuttering onset in infants who are genetically at risk for stuttering.<sup>63,73</sup> If that were true, it would exclude the possibility that disrupted neural speech processing is an effect of stuttering rather than part of its cause. There is indeed evidence of such disruptions in children who stutter, as noted in Lecture Two. There is even some preliminary evidence—unreplicated and involving few participants—that structural brain issues are present in neonates at risk of stuttering.<sup>74</sup>



The developers of the model also venture that their proposition “would be falsified if stuttering were shown to occur during nonsyllabized vocalization, for example during the production of extended vowels” (p. 359).<sup>63</sup> Also, the model would be challenged if recurring findings emerged showing that stuttering did not decrease under experimental conditions that substantively reduced linguistic stress contrasts.

### *Explanatory power*

The P&A Model was designed specifically to explain empirical findings about stuttering, so not surprisingly, it does this well. Its original development was intended to explain that vowel duration variability decreases with treatments that incorporate a fluency enhancing condition.<sup>75,76</sup> The model can explain those treatments in terms of reduced vowel duration variability, which compensates for speech motor system instability by reducing linguistic stress during speech.

The model can also explain the prominence of repeated movements at stuttering onset: as early stuttering moments are triggered by the child’s beginning use of linguistic stress contrasts, the child attempts to stabilise the speech system by minimising linguistic stress contrasts. Subsequent development of idiosyncratic fixed postures and superfluous behaviours are a less adaptive response to the problem. If myelination is involved in the disrupted neural speech processing of stuttering (see Lecture Two), the P&A Model can explain natural recovery for some children and a lifetime of stuttering for others.

The model explains much of the influence of spoken language on stuttering because stuttering on initial sounds and initial words of utterances is associated with linguistic stress. It can also explain why stuttering is more likely to occur on linguistically complex utterances than simple utterances; linguistically complex utterances contribute to instability of speech movements.<sup>77</sup> The threshold factor of the model, involving anxiety and day-to-day dual tasking, can explain differences in stuttering severity across situations and within individuals.

## **The Demands and Capacities Model**

### *Background*

In short, the Demands and Capacities Model states that stuttering is caused by the interaction of many

\* Adapted and reproduced with permission: Packman, A. (2012). Theory and therapy in therapy: A complex relationship. *Journal of Fluency Disorders*, 37, 225–233. © 2012 Elsevier.

factors to be found in the living environments of early childhood, and within children themselves. There is nothing necessarily pathological or atypical about the factors involved. They just interact uniquely for each child to be responsible for stuttering. To say it precisely, the model specifies no causal factors that are *necessary and sufficient* for stuttering development. In the words of its developers, “there is no single etiology, but as many etiologies as there are stories of stuttering development” (p. 24).<sup>78</sup>

As discussed earlier, the Diagnosogenic Theory was clinically influential during the middle of the last century. The Demands and Capacities model seems to be transparently related to the Diagnosogenic Theory, and perhaps that is one reason why it has been clinically influential during the latter part of the last century and during this century.

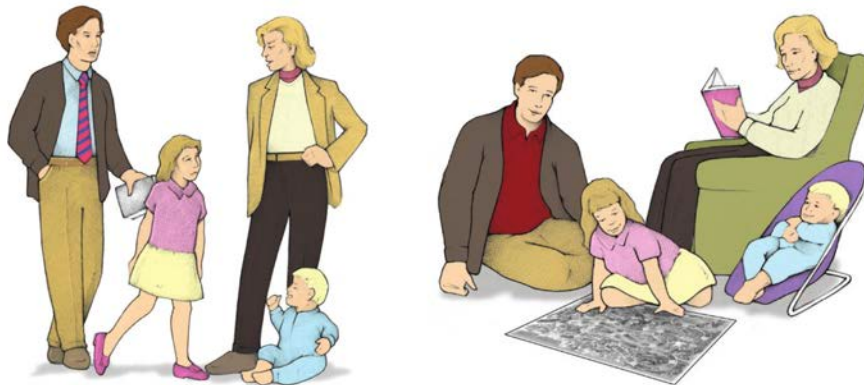
### Overview

The Demands and Capacities Model, as its name implies, states broadly that stuttering occurs when the demands for a child’s fluency exceed the child’s capacity to produce it. The demands on children come from the living environment and include excessive parent language expectations, constant time pressures of living, and excessive parent demands for advanced cognitive performance. Four capacities of children are implicated: speech motor control, language development, social and emotional functioning, and cognitive development

The prominence of the Demands and Capacities Model prompted an entire issue of the *Journal of Fluency Disorders* to be devoted to it in 2000. The model has been described at many sources,<sup>78</sup> and variants have been developed.<sup>79,80,81</sup> The first appearance of the Demands and Capacities model appears to be in a 1987 textbook:

this growing capacity to talk more easily is paralleled by increasing demands for fluent speech, demands placed on children by the people they communicate with ... when the child’s capacity of fluency exceeds the demands, the child will talk fluently but when the child lacks the capacity to meet demands for fluency, stuttering will occur. (p. 75)<sup>82</sup>

The model has been depicted graphically as shown in these figures.\* The situation on the left is where demands exceed capacity, and hence where stuttering occurs. The situation on the right is where capacities exceed demands, and hence stuttering does not occur.



\* Reproduced with permission: Guitar, B (2014). *Stuttering: An integrated approach to its nature and treatment* (4th ed.). Baltimore, MD, Lippincott Williams & Wilkins. © 2014 Lippincott Williams & Wilkins.

*Testability and explanatory power*

Although extensively popular theoretically and clinically, the Demands and Capacities Model has been criticised many times.<sup>4,60,83</sup> Those criticisms reiterate the point that it is not testable and hence not falsifiable. This is for the simple reason that, as quoted earlier, “there is no single etiology, but as many etiologies as there are stories of stuttering development” (p. 24).<sup>78</sup> It is logically impossible to disprove an indefinite number of causes. And because the model specifies no causal factors as *necessary and sufficient* for stuttering development, it is not suited to explain the empirically established features of stuttering.

**Other clinical multifactorial models**

Although there is no treatment that is based directly on the P&A Model, there are two variants of the Demands and Capacities Model on which treatment methods for early stuttering are based. These are now overviewed.

*A family-focused treatment approach*

The first figure\* is a graphic from the Stuttering Center of Western Pennsylvania<sup>84</sup> that depicts a family-focused treatment approach<sup>85</sup> The conceptual similarity between this and the Demands and Capacities Model is apparent, as is the notion that nothing is *necessary or sufficient* for stuttering to occur, as shown by the phrase “factors potentially associated with childhood stuttering.”

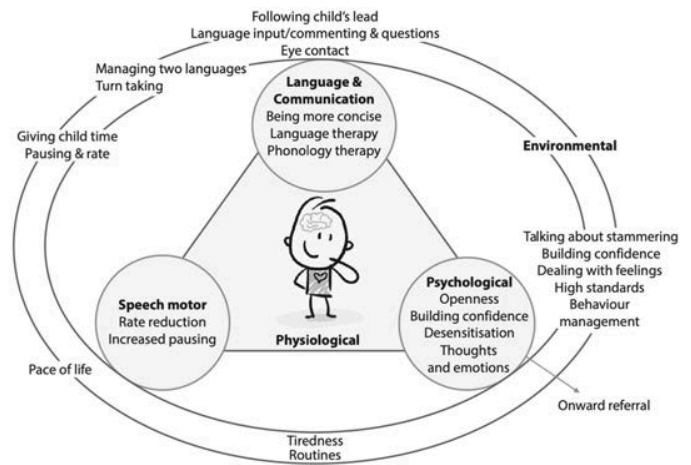
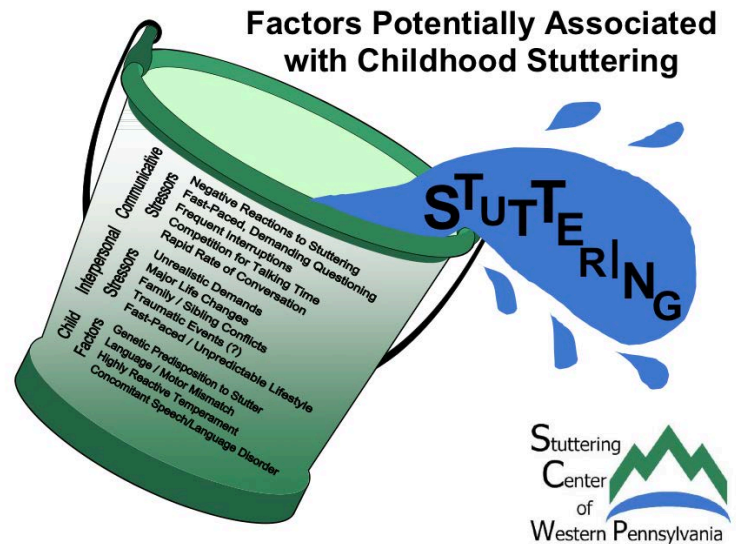
*The Michael Palin Centre Model*

The Michael Palin Centre in London proffers another clinical variant of multifactorial models,<sup>86</sup> which is shown in the next figure.† The conceptual similarity with other multifactorial models is apparent.

**The Erasmus Clinical Model of the Onset and Development of Stuttering 2.0**

The P&A model explains the occurrence of stuttering moments, and the Demands and Capacities model explains causal factors responsible for early stuttering. However, the Erasmus Clinical Model<sup>87</sup> is much broader in scope than either of those; it describes the developmental course of stuttering spanning the period from before birth to adulthood. It might be described as a combined causal model, assessment tool, and clinical case record form. As described by the authors, the purposes of the model are to:

- measure stuttering using scales provided for severity and impact of stuttering



\* Reproduced with permission: the Stuttering Centre of Western Pennsylvania. © 2004 Stuttering Centre of Western Pennsylvania.

† Adapted and reproduced with permission: Kelman, E., & Nicholas, A. (2020). *Palin parent-child interaction therapy for early childhood stammering*. Routledge. © 2024 Routledge.

- provide a means to discuss stuttering development “with clients and parents in an open manner that can be understood by non-experts”
- describe two developmental pathways, “one showing transient stuttering and the other stuttering persistence”
- provide a clinical tool for “discussing developmental processes, assessment results, and treatment options and goals”
- “create and adjust on an ongoing basis a highly individualized Lifeline throughout treatment based on the unique experiences of each individual client” (p. 14).

The authors present their model with a background section that outlines their judgement about research findings that are pertinent to the developmental course of stuttering, along with an overview of six causal theories that influenced their model.

The module is depicted in Figure One (p. 10) and involves “core factors” of genetic predisposition and brain development. These are influenced by “sensorimotor capacity” as it interacts with “influencing factors” of “speech/language/cognitive development,” “speech motor skills,” “temperament,” and “other specific factors” (p. 10). These lead to stuttering that lies on a continuum between “transient stuttering” and “persistent stuttering.” The location of stuttering on that continuum is depicted to involve an interaction between “maturation/growth” that is described as an interaction with a set of variables categorised as “biological factors,” “psychological factors” and “social factors” (p. 10). A case study of this model being used is shown in Figure Two (p. 12), which depicts stuttering throughout life from before birth to adulthood.

The model is presented as something that could be easily understood by clients. Perhaps then, for a tool intended for such a practical application, clinicians would benefit from future research that documents its clinical usefulness.

## The current clinical influence of multifactorial models

### *Clinical research*

Consistent with the current clinical popularity of multifactorial models, they have prompted clinical research. That research has included clinical trials, along with basic research that has explored their clinical value. That basic research has dealt with parent speech rate reduction, pausing when speaking to children (inter-turn speaker latency), parent question-asking, and parent sentence recasts. That body of research is overviewed in Lecture Seven.

### *A study of clinical practices*

In a 2022 survey<sup>88</sup> of 121 Norwegian speech-language pathologists, they were asked: “The first time you meet the parent(s) of a preschool child (0–6 years of age) who is reported to be stuttering, what are the three pieces of advice you give most often?” (p. 941). From a list of 14 options, 78% of the clinicians chose “time to talk,” 51% chose “make eye contact,” 39% chose “increase pauses,” 23% chose “turn-taking,” and 16% chose “reduce activity.”

### *A clinical component checklist of multifactorial models*

Researchers at the Vanderbilt University Medical Centre developed an 18-item scale to assess how parents believe they are implementing treatment components of multifactorial models: The Vanderbilt Responses to Your Child’s Speech rating scale.<sup>89</sup> The scale deals with five factors: “(a) Requesting Change, (b) Speaking for the Child, (c) Supporting Communication, (d) Slowing and Simplifying, and (e) Responding Emotionally” (p. 4652).

## OTHER MODELS OF STUTTERING CAUSALITY

There are many causal explanations of stuttering that emerged in the latter part of the last century that are not multifactorial in the sense that they do not assemble a causal explanation from several contributing factors. Most of these have attracted little attention during this century, according to

whether they have featured in recent peer-reviewed journals or published conference proceedings. Arguably, such explanations are potentially destined to join those with an historical place in the discipline. They include the Sensory-Motor Modelling Theory,<sup>90</sup> the Neuroscience Model,<sup>91</sup> the Anticipatory Struggle Hypothesis,<sup>92</sup> the Two-Factor Theory,<sup>93</sup> the Neuropsycholinguistic Theory,<sup>94</sup> and the Suprasegmental Sentence Plan Alignment Model. Those causal explanations have been reviewed in several reference texts.<sup>8,9</sup>

There has been one causal explanation proposed this century<sup>95</sup> which might generate future interest. However, toward the end of the last century, there were three causal explanations that have generated considerable interest. These focus on one class of factor: the Interhemispheric Interference Model, focussing on the brain, and the Covert Repair Hypothesis and the EXPLAN theory, that focus on motor planning for articulation. These are now overviewed.

## The Interhemispheric Interference Model

### Overview

The broad notion underlying this model has a long history. It dates back to the early years of the last century to Lee Edward Travis<sup>†</sup> who proposed the Orton-Travis theory of cerebral dominance,<sup>99</sup> the origins of which are apparent in a 1925 report about dyslexia.<sup>96</sup> In 1986, just before his death, Travis asserted “the stutterer differs significantly from the normal speaker only in his neuro-anatomical organization for speaking” (p. 119).<sup>97</sup>

It appears that the first formal proposal of the Interhemispheric Interference Model, which implicates the supplementary motor area, occurred in 1987.<sup>98</sup> The model has two parts. The first is that the supplementary motor area of the brain is inefficient, and the second is that the system of hemispheric activation is over-reactive. These two factors are proposed as *necessary* and *sufficient* for the development of stuttering; either factor alone is not necessary. The Interhemispheric Interference Model is an extension of the now defunct Orton-Travis Theory,<sup>99</sup> but departs from it by specifying that those who stutter have typical lateralisation of speech functions. The most recent iteration of the model states, “an anomaly in interhemispheric relations and a deficit in the mechanisms of speech–motor control are each a necessary but not sufficient condition for stuttering” (pp. 125–126).<sup>100</sup>

### Testability and explanatory power

Packman and Attanasio<sup>4</sup> contend that the model is parsimonious because it specifies only two brain factors that are *necessary* and *sufficient* for stuttering. However, they note that it would be difficult to falsify. They draw attention to research findings—up to 2017—that support the model, and some that challenge it. They outline some limitations of the explanatory power for this model.

## The Covert Repair Hypothesis

### Overview

The Covert Repair Hypothesis draws on Levelt’s well-known model of speech production,<sup>101,102</sup> and another model of phonological coding.<sup>103</sup> Levelt’s model, in short, comprises three linear processes. The first is the selection of a lexical concept to be spoken. The second is the selection of a word in abstract form (lemma) and its grammatical encoding. Finally, a “mental syllabary” is accessed<sup>104</sup> and the word becomes a set of syllables ready for articulation.

The central proposition with the Covert Repair Hypothesis is that those who stutter have phonological coding errors in the process of preparation for articulation, and that stuttering moments are covert attempts by the speaker to correct these errors before speech execution of the faulty plan.<sup>105,106</sup> Those who stutter have more errors than those who do not, and consequently they need to correct the errors more. Those corrections occur before the articulatory sequence occurs, and this leads to repeated

<sup>†</sup> Lee Edward Travis is credited as the originator of the speech-language pathology discipline at the University of Iowa, before Wendell Johnson arrived there.

movements and fixed postures during speech.

The hypothesis does not state that there is anything qualitatively different between those who stutter and those who don't, merely that the former group have slower phonological coding and have more errors in the phonetic plan and, hence, they need to make more corrections. In effect, the hypothesis proposes that stuttering and typical disfluency are on either ends of a continuum. As mentioned during Lecture One, this is known as the Continuity Hypothesis.<sup>107</sup>

#### *Testability and explanatory power*

The Covert Repair Hypothesis has attracted considerable research. Overall, some findings support the model, and others falsify it. This body of research, up to 2017, is reviewed by Packman and Attanasio.<sup>4</sup> Those authors outline some limitations of the explanatory power for this causal explanation.

## The EXPLAN Theory

### *Overview*

The EXPLAN theory has in common with the Covert Repair Hypothesis that it draws on Levelt's speech processing model. However, it differs from the Covert Repair Hypothesis in proposing that the motor plan is delayed, rather than being incorrect. The theory seems to be foreshadowed in reports from the late 1990s.<sup>108,109,110</sup> The impetus for its development seems to be that stuttering generally tends to occur more often on content than function words (see Lecture One). The authors proposed a hypothesis that the "stuttering of function words is caused by unavailability of instructions for the following content word" (p. 1020).<sup>108</sup> They also propose that, compared to function words,

the speech plan of a content word is unavailable because planning of such words is relatively slow because of their more complex semantic content, their phonetic composition, and their greater length when compared to function words. (p. 1028)<sup>108</sup>

It appears that the first formal statements of the theory occurred some years later,<sup>111,112</sup> introducing the term EXPLAN theory to capture the fact that it deals with speech planning and execution. Probably the most comprehensive, formal outline of the theory was presented in 2004.<sup>113</sup> The theory deals with the planning of speech as the linguistic aspect of the process, and execution of speech as the motor component. Stuttering occurs when the motor plan is late in presentation for speech execution. According to the theory, this occurs because planning of the linguistic segments of content words is slow; they are more difficult to plan than function words. The theory suggests that stuttering occurs when the speaker uses whole-word repetitions to delay the execution of a motor plan for a content word that is not yet ready. Or the speaker abandons that delaying strategy and instead attempts to progress to speak the incompletely prepared word, hoping that the plan for it will arrive in time. This causes other, more complicated speech perturbations.

As with the Covert Repair Hypothesis, the EXPLAN Theory incorporates the Continuity Hypothesis, linking the typical disfluency of early childhood to stuttering development. This argument is stated clearly in one publication, and draws on the notion that "young speakers, whether they are diagnosed as stutters or not, would exhibit similar nonfluencies" (p. 346).<sup>110</sup> It is the shift during adulthood from disfluencies on function words to disfluencies on content words, for which there is a delayed motor plan, that is responsible for persistent stuttering. Early onset stuttering is the simple repetition of function words, to delay things, because the content word is not ready. Persistent stuttering in adulthood is when the speaker essentially abandons the delaying tactic with function words and attempts to move forward with the content words that are not fully planned, resulting in different, more complicated stuttering moments.

### *Testability and explanatory power*

Packman and Attanasio<sup>4</sup> contend that this theory is testable, and outline research until 2007 by independent groups that supported and refuted it. Since then, another independent group<sup>114</sup> conducted



two experiments with adults, using a “semantic blocking” research protocol, and reported results consistent with the theory. Packman and Attanasio<sup>4</sup> state that “it is parsimonious in that it posits one factor as the cause of stuttering” (p. 124).

### **SUMMARY AND CONCLUDING COMMENTS**

The cause of stuttering is a necessary topic for discussion with clients and parents. The viability of a causal explanation includes its testability and explanatory power. Causal explanations of stuttering can be classified as those that are multifactorial, assembling a causal explanation from several contributing factors, and those that are not. Some multifactorial explanations of stuttering have immediate clinical application in the sense that treatments are based on them. Outlines of those treatments and clinical trials of them are considered in Lectures Six and Seven. There is much yet to be known about causal factors of stuttering, but for now sufficient is known to state the following, which is useful for clinical practice. Stuttering is a genetically influenced condition involving atypical neural speech processing that manifests at an early stage of language development and may be significantly moderated by cognitive factors thereafter, commonly social anxiety.

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## LECTURE FOUR: CLINICAL MEASUREMENT OF STUTTERING<sup>†</sup>

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### SIX REASONS FOR CLINICAL MEASUREMENT

#### Assessment

Clinical measurement provides a formal way to document the nature and severity of the impact of stuttering for clients when they first come to a clinic. Such impact may be behavioural or non-behavioural. Much of the non-behavioural impact of stuttering will be related to anxiety, which is outlined during Lecture Ten. Measurement of anxiety is described during Lecture Eleven. The behavioural impact of stuttering will be related to stuttering moments and how often they occur, as described during Lecture One.

This does not mean that clinical measurement is necessary to detect the impact that stuttering has for clients. To the contrary, with clinical experience, that impact will be obvious. However, clinical measurement provides numbers that quantify the impact of stuttering. For many reasons, it is a useful thing to record these numbers during client assessment. There are reference texts<sup>1,2</sup> that provide an overview of most formal clinical assessments for stuttering adults and children. Many of these assessments are not discussed here because they are used mostly in research contexts, not clinical contexts.

There is evidence that many of the assessments considered in this lecture can be done with video telehealth as well as in-clinic. A study of telehealth assessments with 14 adult stutterers<sup>3</sup> gave seven of them an in-clinic assessment and seven of them a video telehealth assessment. The 70-minute assessments were done by speech-language pathology students and involved interview and formal testing procedures. Results showed that the two assessment methods were comparable in terms of time required, assessment results, and the client experiences of assessment. A study of 30 Arabic child stutterers,<sup>4</sup> ages 6–15 years, gave half an in-clinic assessment and half a video telehealth assessment. Three measures considered during this lecture were used (percentage syllables stuttered, a severity rating scale, and the Stuttering Severity Instrument). The authors concluded that video telehealth assessment was “feasible, reliable, and valid,” and was associated with “high levels of satisfaction” (p. 12) recorded by children and parents.

#### Communicating with clients

Clinical measures establish a common language between clinicians and clients, or parents, that can be used to communicate easily about everyday stuttering severity. For example, if a parent stated of a child “he was a 5 all yesterday,” the clinician would immediately have a clear picture of the child’s stuttering severity during that day. Such communication between clinician and client is essential to assess whether treatments are working as planned.

#### Stating treatment goals

When clinicians give stuttering treatments, they need a clear idea of what they are intending to achieve. Clinical stuttering measures can be used to convey to clients, or their parents, what those intended achievements are. For example, a clinician may say to a client: “That test shows that the impact of your stuttering is much less than six months ago.” The formal description of this process uses terms such as *setting of treatment targets*, *treatment target criteria*, or *treatment goals*. Some treatments have standard, built-in treatment target criteria that may not be advisable to change, and for other treatments it is usual for the clinician and client to determine the treatment goals together.

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<sup>†</sup> Thanks to Sue O’Brian for guidance with this material. This lecture deals with clinical measurement of stuttering severity, impact of stuttering, and speech satisfaction for those who stutter. Measures of anxiety are discussed during Lecture Eleven (Treatment of Social Anxiety and Stuttering).



Using measurement to document treatment goals and whether they have been met is part of treatment accountability. Stuttering treatment must be funded, either through government or private sources. Those who provide that funding—government health care providers or the clients—need to know the outcome of their investment. Clinical measurement is an ideal way to provide that accountability by documenting client health improvements and how many hours of funded treatment were required to attain those improvements.

### Assessing progress toward treatment goals

Clinical measurement does not stop after assessment. It is necessary to determine if a treatment is working as planned and that satisfactory progress is being made towards treatment goals. If progress towards those goals is not satisfactory, clinical measures may assist with documenting and exploring why that is the case, so that the problem can be fixed.



### Managing maintenance of treatment gains

Health care resources for stuttering are valuable. Consequently, those resources are used inefficiently if clients do not maintain their treatment gains, and if they return to the clinic for more treatment, perhaps several times, after their original treatment. As will be discussed in Lecture Ten, such post-treatment relapse is a serious problem with adult clients. Post-treatment relapse is not so much of a problem with young children who stutter who successfully complete treatment, but it does occur.<sup>5</sup>

Clinical measurement can be used to monitor client post-treatment progress to detect any signs of impending relapse and to provide a clinical response if it begins to occur. The period after treatment that is designed to prevent relapse is referred to as *maintenance*. It is an indispensable part of any stuttering treatment.

### Keeping track of daily stuttering severity changes

As noted in Lecture Three, stuttering severity is notoriously variable. To reiterate, stuttering severity is likely to vary with how many people are being spoken to at one time, with usually more stuttering when there are larger audiences. Stuttering will typically change severity across everyday situations, with lower severity typical of familiar conversation partners, and more severe stuttering likely when speaking with formal acquaintances and figures of authority. It is essential for clinicians to use clinical measurement to know about and keep track of such day-to-day variations during clinical management. For example, a clinician might ask a client to use a technique to control stuttering in a daily situation in which severe stuttering typically occurs. The clinician may ask the client to measure stuttering severity in that situation each day to explore whether systematic improvement is occurring.

## SEVERITY RATING (SR) SCALES

### Overview

#### *Differing numbers of scale divisions*

Severity ratings are perceptual measures, where an observer listens to a sample of stuttered speech and uses the severity rating SR scale to record an overall judgement of severity. Or a client can self-assign a SR score.

Severity rating scales have been around for years in various forms. They vary according to how many scale divisions there are, but the number is arbitrary. There is no real reason to think that a certain number of divisions is better than any other. Seven-point scales, 9-point scales, 10-point scales, and

11-point scales are commonly used.<sup>†</sup> Often, but not always, some or all the scale divisions have labels telling the user what they represent.

#### Commonly used clinical severity rating scales

An example of a severity scale used for research is used in the Illinois Early Childhood Stuttering Project (see Lecture Two). One version of the scale is 0 = *normal disfluency*, 1 = *very mild stuttering*, and 7 = *very severe*,<sup>6</sup> and another version is 0 = *normal speech*, 1 = *very mild stuttering*, and 7 = *very severe stuttering*.<sup>7</sup> A scale commonly used during treatment of early childhood stuttering (see Lecture Six) is 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 10 = *extremely severe stuttering*.

#### Presenting severity rating scales to clients

It is useful to present SR scales visually to clients and parents, as well as describing them, and for them to have a copy in some form for their use outside the clinic. Here is how a SR scale might look when presented to clients:



#### Equal interval ordinal scales

SR scales are called equal interval ordinal scales. “Ordinal” means a sequence of numbers, and “equal interval” means it is intended that each scale division represents the same severity increment.

Whether, in practice, such scales really are equal interval scales, or whether people tend to bunch up scores somewhere on them, is a complicated matter of psychophysical analysis.<sup>8</sup>

#### Reliability of severity ratings

By 2011, there were 11 research reports about SR reliability for stuttering as indicated in a publication (see Table 1, p. 1287).<sup>33</sup> For children, there is some evidence that, with little training, clinicians and lay listeners agree when using an SR scale,<sup>9</sup> and that parents of children who stutter have close rating agreement with clinicians.<sup>10,11</sup> One report<sup>12</sup> used a 10-point SR scale with 3–6-year-olds and seven languages, and clinicians who spoke those languages: Danish, English, French, German, Greek, Italian, and Persian. Results showed that neither language nor clinical experience influenced the clinician ratings.

For adults, one report<sup>13</sup> showed that clinicians do not use the scale reliably when adult clients speak an unfamiliar language of Mandarin. A study of 25 English speaking clinicians,<sup>49</sup> who did not speak Spanish, showed adequate relative reliability with a 9-point SR scale when it was used for Spanish speech samples. However, there was a problem with absolute reliability among the clinicians, leading the authors to caution about the clinical generalisability of the scale into another language. A study of 195 participants<sup>14</sup> 172 of whom were adults or adolescents, used clinician severity ratings in real time after a 5-minute conversation: 1 = *no stuttering at all*, 2 = *extremely mild stuttering*, and 10 = *extremely severe stuttering*. An independent clinician used the 10-point scale to measure stuttering severing from video recordings of the conversations. There was a strong correlation of  $r = .68$  between the two sets of recordings, and 76% of ratings were within one scale value of the original rating.

<sup>†</sup> An example of an 11-point scale is 0–10.

### A severity rating scoring guide<sup>†</sup>

After listening to a client for whatever period seems reasonable to be a valid speech sample, these four questions can be used to guide the assigning of a SR using a scale where 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 10 = *extremely severe stuttering*.

*Were there any unambiguous stuttering moments?*

If not, then the score is SR 0, which means *no stuttering*. If there were some ambiguous stuttering moments that could have been typical disfluency or might have been stuttering, then SR 1 would be appropriate, meaning *extremely mild stuttering*. Also, SR 1 would be appropriate if there was one unambiguous stuttering moment that was brief but not particularly bothersome: perhaps a syllable that was quickly repeated two or three times without a fixed posture or superfluous behaviour. Possibly, SR 2 might be appropriate in that situation, particularly if there was more than one such brief stuttering moment, indicating a little more severity than *extremely mild stuttering*.

*Would a casual observer notice the stuttering?*

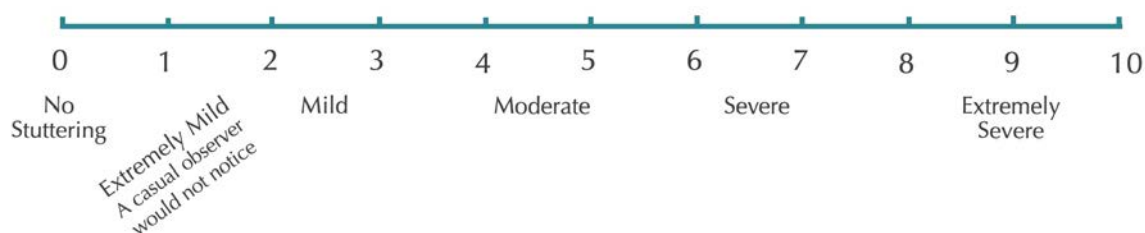
A rule of thumb is that a casual listener would not normally notice SR 0–1: someone without a speech-language pathology background who would not make a sophisticated judgement about speech. That would be someone from the public who the client might encounter during everyday life, such as an accountant, bus driver, shopkeeper, waiter, lawyer, and so on. If it seems that such a casual observer would notice the stuttering, the SR would be 2 or more.

*How much does it affect communication?*

As discussed during Lecture One, stuttering is time consuming and on average those who stutter can say one third less than those who do not stutter, and with severe stuttering speech output might be less than a quarter than that of peers. So, a prime consideration when assigning a SR score is the extent to which stuttering affects communication. Reduced speech output will be one part of that consideration, as will how socially distracting stuttering may be because of superfluous behaviours. In cases where superfluous behaviours are particularly socially distracting and time consuming, a clinician may feel that communication is particularly affected.

*Was it mild, moderate, or severe?*

It is useful to think of five categories to describe how stuttering affects communication: *extremely mild*, *mild*, *moderate*, *severe*, or *extremely severe*, as shown below.



### The clinical population as reference

Severity rating scales are of most use clinically if they are assigned with reference to the clinical population of those who stutter. In other words, SR 5 means a client is similar to others who stutter and come to clinics with that stuttering severity. At present, the only way to establish such reference points is with clinical experience or with mentoring from a senior colleague. There are no generally available training methods to show inexperienced clinicians what a group of experienced clinicians

<sup>†</sup> Thanks to Sue O'Brian for this material.

believe are representative SR scores for the clinical population. Ideally, clients and clinicians would all give the same SR for the same speech sample. In practice, though, experienced clinicians generally accept one-unit margins as acceptable limits, such as SR 6–7, SR 4–5 and SR 7–8.

Clinical knowledge about the severity of the general population can be used to guide clients when using the SR scale. For example, if a client says that speech during a conversation in the clinic was SR 7, the clinician might say, “that was more like a 5.” After watching a parent and child talking for a while, a clinician could ask “what SR would you give that speech just then?” The parent might say “4,” and the clinician might say “yes, I agree,” or the parent might say “3,” and the clinician might say, “I would have given that a 4.” Most clients and parents quickly learn to match the SR that the clinician would give.

### Advantages of severity ratings

#### *Simple*

An advantage of SRs is that they are simple and require no equipment. Additionally, it seems that extensive training is not needed to learn to use them,<sup>9,10,11</sup> so they are particularly suitable for clients and parents, who can use the measures with themselves or their children. Further, they can be used easily with other languages.<sup>15</sup>

This means that clinicians can have direct access to information about how severe stuttering has been during a certain period. For example, a clinician might say to a client “how was your stuttering last week” and receive a reply “1.” In which case, the clinician knows that the client’s stuttering was extremely mild during the previous week. Another example question would be “how has your stuttering been during phone calls to that customer?” The simplicity of the SR scale allows it to be used with clinical flexibility. For example, a clinician may ask a client or parent to record a “typical SR” and a “worst SR” during a defined period during a day, or for an entire day.

#### *Valid*

It seems that SRs are valid because they take account of all behavioural features of stuttering rather than just stuttering moments. Also, the study of 195 participants mentioned earlier<sup>14</sup> showed strong correlations between self-report and clinician ratings using the 9-point severity rating scale.

#### *Covert*

Severity ratings can be done covertly. Clinicians can assess clients’ stuttering severity in the clinic without them being aware it is occurring. This prevents the so-called Hawthorne Effect with stuttering assessment, where behaviour can change when it is overtly assessed. In the case of children, parents can use SRs to covertly measure their children’s stuttering at any time of the day when they are together.

### An example of severity ratings during treatment

Appendix One to this lecture is an example of a clinical file showing the use SR measures during treatment of a child for 12 weeks. The child scored SR 0 consistently for the last few weeks of the file record, with only the occasional SR 1. This is an example of a successful treatment of childhood stuttering. The parent SR scores are indicating 0 = *no stuttering* most of the time, with the occasional SR 1 = *extremely mild stuttering*, which a casual observer would probably not notice.

## PERCENTAGE SYLLABLES STUTTERED (%SS)

### Overview

The views of 12 scholars in the field about stuttering assessment<sup>16</sup> included a core component of “speech fluency and stuttering behaviors” (p. 2379), but there was no universally agreed method for such assessment. Percentage syllables stuttered, commonly abbreviated to %SS, is one such assessment. Compared to severity rating scales, %SS is not a straightforward procedure; it requires

equipment, and it is more logistically and arithmetically complicated, and clinically challenging. As such, clinicians may choose only to understand %SS for the purpose of reading clinical research literature, in which it features prominently. They may prefer not to use it in the clinic.

Percentage syllables stuttered is a measure of the percentage of spoken syllables that are stuttered. It is sometimes referred to as a stutter-count measure because it is based on a count of unambiguous stuttering moments. To reiterate from Lecture One, unambiguous stuttering moments are moments during speech that are clearly stuttering and not typical disfluency.

Percentage syllables stuttered is based on syllables spoken, the syllable being a fundamental unit of speech production.<sup>17</sup> The average number of syllables in each word spoken increases from childhood to adulthood as language complexity develops. During adulthood the ratio is around 1.5 syllables per word spoken, but during the early years of life the ratio is much lower at 1.15, according to one source.<sup>18</sup>

When measuring %SS, syllables are thought of as being stuttered or not stuttered. For example, if someone speaks 900 syllables and 98 of them are unambiguous stuttering moments, that is 10.1 %SS. If someone speaks 1,435 syllables and 75 of them are unambiguous stuttering moments, that is 5.2 %SS. Percentage syllables stuttered is usually written to one decimal place.

### How percentage syllables stuttered is measured

A study explored the relative merits of standard and “challenge” phone calls to assess %SS.<sup>19</sup> The latter calls involved occasionally (but courteously) interrupting, disagreeing with, and talking over participants. Results showed little difference between the two approaches in terms of statistical analysis. However, the challenge phone calls elevated participant anxiety slightly, and for some participants this resulted in clinically significant increases of stuttering severity. Hence, the researchers argued that, in clinical practice, challenge phone calls might be a more valid speech assessment than standard phone calls.

There is evidence that, for adults, a %SS score during a 10-minute everyday conversation is representative of stuttering severity during that entire day.<sup>20</sup> That finding has been replicated with adolescents,<sup>21</sup> with the caveat that the finding pertains to group data only, but not to individuals. The former of those studies<sup>20</sup> is useful when interpreting %SS scores clinically. The speech of 10 adult participants was studied continuously during a 12-hour day, during which time their mean number of syllables spoken was 33,617, with a range of 17,274–50,463, and a standard deviation of 9,027 syllables. So that means, for example, if an adult stutters at 10 %SS for a 12-hour day, there could be somewhere between 1,700 to 5,000 stuttering moments during that day. Such data are currently not available for children.

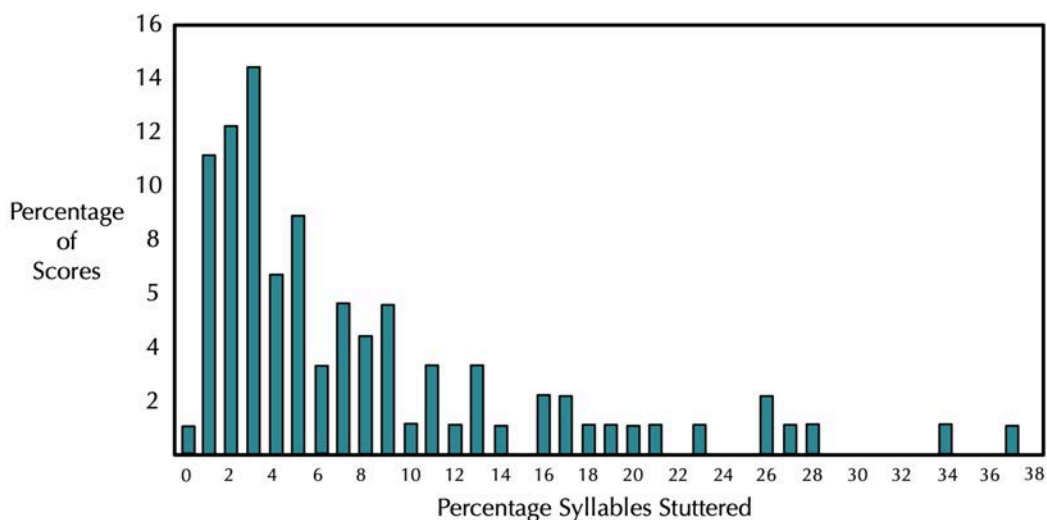
When calculating %SS, a syllable is counted as stuttered only once, regardless of how many stuttering behaviours are associated with it. For example, consider “yesterday I-I-I-I, you see I, you see I, well, um I-I-I-I was here.” That is counted as six syllables—“yesterday I was here”—with one of them stuttered. The fact that there were repeated movements and superfluous verbal behaviours with saying “I,” and two attempts to say it, does not change the fact that, for the purposes of calculating %SS, it was just one stuttered syllable.

Less commonly, percentage words stuttered is used, which is a similar measure but calculated by counting every word spoken, not every syllable spoken. That measure is now used rarely for research and clinical practice.

It is sometimes claimed that %SS is an objective measure, but pedantically speaking, that is not correct. When measuring stuttering with %SS an observer needs to make a perceptual judgement about whether a syllable is stuttered or not; there is no objective truth to it. Another pedantic point is that the terms “stuttering frequency” and “stuttering rate” are often used interchangeably when referring to %SS. In a strict sense, however, percentage is a measure of proportion, so those terms are not correct. But they are commonly used, nonetheless. It is more correct to refer to %SS as a measure of stuttering severity.

### Percentage syllables stuttered scores are not normally distributed

The distribution of %SS scores is not normal.<sup>22</sup> There are more mild cases than severe cases.<sup>23,24, 25</sup> Information from the latter report<sup>25</sup> about adults is reproduced in the diagram below.\* The correct way of describing this distribution is to say that it is skewed to the right, and resembles a negative binomial distribution. The situation seems fairly much the same with early childhood stuttering,<sup>26</sup> although %SS scores generally seem to be lower at that time of life.<sup>23</sup> In the figure below there are few people with scores greater than 20 %SS, but many with scores below 10 %SS. The median score is 4.8 %SS. The 60<sup>th</sup> percentile is 6.5 %SS, meaning that 60% of the scores—at least from this data set—are below 6.5 %SS.† For research that involves %SS, the implications of skewed scores are a little complex because there are mathematical issues with how they should be analysed and interpreted. Those statistical issues are discussed in detail elsewhere.<sup>27</sup>



It seems that SR scores are more normal-looking than %SS scores.<sup>22</sup> This is noticeable with clinician-scored SR for adults,<sup>25</sup> and when adults apply SR to themselves, one report showed that the result is almost a perfect normal distribution.<sup>28</sup>

### Equipment for percentage syllables stuttered measurement

During clinical practice, %SS is typically measured during a conversation with the client/child, or while observing a conversation between a child and parent. A two-button counting device is used for measuring %SS, such as the one in the figure. One button is pushed for every syllable spoken without stuttering and the other button is pushed for every syllable spoken with unambiguous stuttering. The device automatically calculates %SS. Commercially available devices or smartphone applications can be used, or software is available for laptops.<sup>29,30</sup> Considerable training is required to learn to use such equipment.



\* Adapted and reproduced with permission: O'Brien, S., et al. (2004). Measurement of stuttering in adults: Comparison of stuttering-rate and severity-scaling methods. *Journal of Speech, Language, and Hearing Research*, 47, 1081–1087. © 2004 American Speech-Language-Hearing Association.

† The %SS scores in the figure are rounded to the nearest whole value. That is why there is one case of a zero score; the actual score was 0.3 %SS.

## Limitations of percentage syllables stuttered

### *Validity*

Counting the number of stuttering moments is not necessarily a valid reflection of how severe stuttering appears to an observer. This is because some stuttering behaviours may seem to be more severe than others. For example, fixed postures may appear to be more severe than repeated movements. Regardless, as discussed below, generally there is a strong and clinically significant correlation between %SS and perceptual measures of stuttering severity. But, of course, that will not necessarily be the case for any individual client.

### *Not viable for self-assessment*

It is useful if clients and parents can self-administer a stuttering severity measure during everyday life. But considering the equipment and training needed for measuring %SS, this is not a clinical option. Consequently, any clinical use of %SS is normally constrained to measurement in the clinic by the clinician. An option to obtain %SS measurements of clients during everyday speech is to have them make audio recordings—or even video recordings—of themselves, or have parents make recordings of their children, to bring to the clinic. Such recordings can be made easily with phones. Although it is time consuming to measure %SS from such recordings, in some clinical contexts the effort might be justified.

### *Reliability*

*Reliability* is a general term for how well a measure gives the same score when used several times. *Relative reliability* refers to how well a measure rank orders *groups of participants*. It is most often measured with a correlation coefficient or an intraclass correlation index. *Absolute reliability* (sometimes known as *agreement*) refers to the closeness of individual scores to each other and to a hypothetical “true score.” It can be measured with percentage agreement, standard error of measurement, or limits of agreement.

Percentage syllables stuttered is a notoriously unreliable measure. The first report about this was in 1940,<sup>31</sup> showing poor absolute reliability: 20 clinicians ranged from 37–136 counts of stuttering moments from the same speech sample. During subsequent decades consistent evidence of such reliability problems emerged.<sup>32</sup> One paper<sup>33</sup> lists 32 research reports that contain measures of reliability. A recurring problem was absolute reliability: if one observer gives a certain %SS score, there is no guarantee that another observer will give the same score or a similar score.<sup>34,35,36,37</sup>

In the most notorious of these studies,<sup>34</sup> researchers gave the same 10 audio recorded samples, eight of which contained stuttering, to 26 clinicians in four countries and asked for their %SS measures. The clinicians gave entirely different scores. Of particular concern was that scores for some samples in the low range had considerable variation: 0–4.2 %SS, 0.6–3.5 %, 0–2.1 %SS, 0–4.8 %SS, and 0–2.1 %SS. Such results suggest that some clinicians would consider some samples to contain no stuttering at all, while other clinicians would consider that the same samples had clinically significant stuttering that would require treatment. A more recent study<sup>38</sup> showed that students and generalist clinicians recorded less than half the number of stuttering moments as experienced clinicians.

In response to these reliability problems, a time-interval stuttering-count procedure<sup>39</sup> was adapted for stuttering.<sup>40,41,42</sup> With this method, the observer notes whether short periods of speech, such as 10 seconds, are stutter-free or whether they contain one or more stuttering moments. However, a subsequent review<sup>43</sup> showed that this method did not solve the reliability problems with stuttering-count measures.

It also appears that %SS reliability problems cannot be solved by listening to speech samples twice and counting syllables the first time and stuttering moments the second time.<sup>44,45</sup> The latter of these studies<sup>45</sup> also reported that it did not help to slow down speech samples while counting syllables and stuttering moments. A report about training procedures for stuttering counts<sup>46</sup> was more encouraging; however, the training by no means solved the reliability problems with stuttering counts. A study with student speech-language pathologists found that they are more reliable when counting stuttering

moments only, compared to counting stuttering moments concurrently with spoken syllables and measuring speech naturalness (see below).<sup>47</sup>

There is evidence<sup>48</sup> that inexperienced observers are more reliable when they indicate whether each utterance contains one or more stuttering moments, compared to when they judge whether each syllable contains a stuttering moment. The same report found that reliability increased when observers had access to transcripts of the speech concerned. The observers were also able to complete the assessment task more rapidly when they had access to transcripts.

There is evidence<sup>49</sup> that %SS training in English does not necessarily generalise to using %SS with another language. Twenty-five English speaking clinicians, who did not speak Spanish, were trained to use %SS. They did not attain relative and absolute reliability when measuring %SS for Spanish speakers.

## THE RELATION BETWEEN SR AND %SS

### A strong relationship

There is a strong correlation of .91 between these two scoring methods when used by clinician observers for the same speech samples.<sup>25</sup> This has been shown also in Kannada (an Indian language).<sup>50</sup> This means that the two measures can be used interchangeably with some confidence, but with two reservations. In one study<sup>25</sup> what prevented a higher correlation were several cases where the %SS score did not correspond at all with the SR score. This occurred several times when samples of stuttering had high proportions of repeated movements or low proportions of fixed postures.

The second reservation about the matter is a report<sup>51</sup> that, to be reliable, %SS scores depend more on audiovisual samples than SR scores. Percentage syllables stuttered scores were 18% higher when scored using audiovisual samples than audio only samples, but this did not occur for SR scores. This is not an issue when talking face-to-face with clients and measuring stuttering severity, but it does suggest that SR is a preferable measure when clients bring audio recordings of their speech to the clinic.

For clinical research purposes, it seems that, at least for early childhood stuttering, %SS and SR do equally well for documenting the results of stuttering treatment during clinical trials, and so the simplicity of SRs makes them a better option in that context.<sup>22,52</sup> (Clinical measurement during clinical trials is discussed during the next lecture.)

There is some evidence that, considering behavioural complexity, clinician severity ratings are a more valid measure than %SS and client self-rated stuttering severity.<sup>53</sup> As outlined during Lecture One, a stuttering moment can involve repeated movements, fixed postures, or superfluous behaviours. The report<sup>53</sup> found that clinician-rated stuttering severity had a significant relationship to the complexity of stuttering moments in terms of how many repeated movements, fixed postures, and superfluous behaviours were present. No such relationship was found for %SS or client-rated stuttering severity. When explaining the latter finding, the authors suggested that, when judging severity, those who stutter focus on the experience of stuttering rather than its behavioural manifestations.

### Repeated movements and fixed postures

Repeated movements are generally not as socially distracting as fixed postures, and they quite often consume less time. So, if a %SS score for a sample is quite high because of many stuttering moments with repeated movements, it will not necessarily mean that the SR score for that sample will be high also. Observers may not think that all the stuttering moments with repeated movements are particularly severe stuttering.

Conversely, consider a speech sample that has a quite low %SS score because there are only a few stuttering moments, but those few stuttering moments are fixed postures, and they are particularly socially distracting and time consuming. Such a sample might score a low %SS but a higher SR because the distracting and time-consuming nature of those fixed postures leads an observer to believe



that stuttering is quite severe.

### Percentile ranks for %SS and SR

The table\* shows the comparative percentile ranks for the two measures for an adult caseload.<sup>25</sup> Clinicians gave both measures based on 3-minute video speech samples. The table shows, for example, that the 50<sup>th</sup> percentile for SR is 3.0 and for %SS is 4.8. In other words, for that data set, half of a clinical caseload will be below those values and half will be above.<sup>†</sup>

PERCENTILE	SR	%SS
10	1.2	1.2
20	1.5	2.0
30	1.9	2.9
40	2.6	3.6
50	3.0	4.8
60	3.9	6.5
70	4.7	8.6
80	5.6	12.6
90	6.8	19.5
100	8.0	36.9

### SYLLABLES PER MINUTE (SPM)

Sometimes a clinical measure of speech rate, mostly syllables per minute, is associated with %SS. Devices that measure %SS typically have a timer that allows syllables per minute measures to be generated. Because stuttering moments consume time, if stuttering decreases after treatment, then speech rate would be expected to increase. It is necessary to use this clinical measure during a treatment that incorporates speech rate targets, which some modern treatments for adolescents and adults do. Progress has been made toward development of a smartphone application for monitoring and feedback of client speech rate.<sup>54</sup>

### SPEECH NATURALNESS (NAT) MEASUREMENT

#### Why measure speech naturalness?

The speech restructuring treatments that figure prominently in these lectures are clinically useful for reducing stuttering but may not produce speech that sounds completely natural. This has been known for a long time to be clinically problematic.<sup>55</sup> Speech restructuring treatments involve a trade-off between speech with reduced stuttering moments and speech that sounds natural. So, a measure of speech naturalness is useful during such treatments to measure how natural clients sound and to guide them in attaining speech that sounds as natural as possible, while providing the desired stuttering reduction.

#### A scale of speech naturalness

A 9-point scale was developed during the 1980s and 1990s and is now used widely by researchers, and sometimes clinicians, to record speech naturalness.<sup>56,57,58,59</sup> For research purposes, it has been shown mathematically that:

For posttreatment data, the average of three independent raters, and for pretreatment data, the average of five independent raters should give a result within one scale point of the hypothetical true score for the speaker in at least 80% of samples. (p. 718)<sup>60</sup>

There is evidence<sup>61</sup> that speech pathology listeners and general community listeners give different scores to clients who are using a speech restructuring technique to control their stuttering. The community listeners gave scores 1.6 scale values higher—more unnatural—than the speech pathology listeners. Also, among the community listeners, men gave scores 1.3 higher—more unnatural—than women.

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† The paper used a 1–9 scale, but the table converts the data to a 0–8 scale.

## STUTTERING-LIKE DISFLUENCIES

Stuttering-Like Disfluencies is a measure that is used for research publications from the Illinois Early Childhood Stuttering Project (see Lecture Two) and has also been used by some other researchers as a measure of stuttering severity.<sup>62</sup>

The three disfluency types most typical of stuttering in young children (part-word repetition, monosyllabic word repetition, disrhythmic phonation) were combined to form a global category that we labelled Stuttering-Like Disfluencies ... (p. 38)

The language of the measure—"stuttering-like"—is ambiguous, and consequently it has been criticised several times because it is not clear to what extent it relates to stuttering or typical disfluency.<sup>63,64,65,66</sup> Part of the issue is that the taxonomy specifies that children who do not stutter have fewer than 3.0 Stuttering-Like Disfluencies per 100 syllables, which implies that nonstuttering children show speech behaviours that can be referred to as "stuttering-like." The potential problem arising from this paradoxical terminology is illustrated in a publication<sup>67</sup> that used the measure and had the following wording in its title: "... frequency of stuttering in young children who do and do not stutter" (p. 2133).

There is a complicated algorithm based on Stuttering-Like Disfluencies: Weighted Stuttering-Like Disfluencies.<sup>68</sup> The algorithm is designed to predict natural recovery from early childhood stuttering. It is derived from coded transcripts of language samples:<sup>69</sup>

The weighted SLD is computed by summing PW [*part-word*] and SS [*single-syllable*] repetitions per 100 syllables of speech and then multiplying this value by the mean number of PW and SS RUs [*repetition units*] combined. This value is added to twice the sum of blocks and prolongations (collectively called as DPs) [*disrhythmic phonations*] (p. 2559) [*italics added*]<sup>70</sup>

## THE STUTTERING SEVERITY INSTRUMENT (SSI-4)

The Stuttering Severity Instrument examiner's manual<sup>71</sup> is now in its fourth edition, often abbreviated to SSI-4. The SSI-4 is a more detailed measure of stuttering severity than either %SS or SR. It involves a composite single-number index that contains information about %SS, the duration of the three longest stuttering moments, verbal and nonverbal superfluous behaviours, and speech naturalness. The speech naturalness scale is the one described above. The superfluous behaviours, referred to as "physical concomitants," are scored on a 6-point scale where 0 = *none* and 5 = *severe and painful looking*. The SSI-4 can be scored manually or with a computerised version. For comparison purposes, there are normative data for 72 young children, 139 school-age children and 60 adults. To use this measure, the forms and manual need to be purchased from the publisher. The test has been translated to Persian.<sup>72</sup>

The SSI-4 is designed for research and clinical applications. It is reported often in stuttering research reports, although not as commonly as %SS. It takes considerable time to complete because client speech needs to be transcribed and analysed. Its time requirements are not an issue for research applications but may be an issue for clinical applications where a stuttering severity measure is required at each weekly appointment.

There have been several reports questioning the reliability of this measurement instrument, which have been reviewed in a more recent report that again questioned its reliability.<sup>73</sup> Another report<sup>74</sup> shows that the SSI-3 (the previous version to SSI-4) provides no additional information than can be obtained from a SR scale. Considering this, and considering that it involves expense to purchase and clinical time to complete, the SSI-4 may not be a useful routine measure for generalist clinicians. However, clinicians who specialise in stuttering treatment may wish to purchase it and commit the time needed to complete the assessment before and after treatment, and perhaps on one or two occasions during treatment.

## THE SPEECH EFFICIENCY SCORE (SES)

A group of researchers has begun developing this measure as an alternative to stutter-count

measures.<sup>75</sup> The SES is derived from waveform analysis of speech, and calculates “the portion of the time during which the speaker produces speech fluency out of the overall speech time” (p. 62).<sup>75</sup> Encouraging results were presented for 15-second audio speech samples, showing that the SES is a viable alternative to SR and %SS. The intention of this research is to eventually to develop “algorithms for automated segmentation and calculation of the SES” (p. 67).

## **THE OVERALL ASSESSMENT OF THE SPEAKER’S EXPERIENCE OF STUTTERING (OASES)**

The impact of stuttering can be measured with the Overall Assessment of the Speaker’s Experience of Stuttering,<sup>76</sup> commonly known as the OASES. It is designed to reflect the World Health Organization’s International Classification of Functioning, Disability, and Health.<sup>77</sup> Each of the OASES questions requires a response on a 5-point scale, with higher scores reflecting more adverse impact.

The OASES is a questionnaire with four categories of questions about the impact of stuttering: general information, reactions to stuttering, communication in daily situations, and quality of life. The OASES score is the total of the four sections. It takes around 20 minutes for the client to complete. The scale was developed in 2006<sup>78</sup> and appears regularly in publications. To use this measure, the forms and manual need to be purchased from the publisher.

Arguably, one useful function of these materials is to guide a clinical interview about the impact of stuttering for clients, based on high-scoring responses to certain items. However, the OASES was designed to be part of a clinician’s assessment tools for documenting impact of stuttering before and after treatment. For this purpose, there are three versions for different ages. The OASES-S is for school-age children 7–12 years, the OASES-T is for adolescents 13–17 years, and the OASES-A is for adults 18 years and older. The OASES-A has been shown to have acceptable reliability and validity,<sup>86,78</sup> and preliminary results for OASES-S and OASES-T are reported in the scoring manual.<sup>76</sup> The OASES-S has been translated into Dutch (OASES-S-D)<sup>79</sup> and Portuguese (OASES-S-PT),<sup>80</sup> and the OASES-A has been translated into Japanese (OASES-A-J),<sup>81</sup> Kannada (OASES-AK),<sup>82</sup> and Hebrew,<sup>83</sup> with data showing it to be reliable and valid in those languages. All OASES versions have been translated to Swedish<sup>84</sup> and Polish<sup>85</sup> and been shown to be reliable and valid. There are OASES Australian normative data to supplement North American normative data.<sup>86</sup> A Simplified Chinese version of the OASES-A<sup>87</sup> suggested that, in China, the impact of stuttering may be greater than in other countries for which data are available. The OASES-A has been shown to capture dimensions involving spontaneity while speaking<sup>88</sup> and feelings of everyday satisfaction with communication.<sup>89</sup> OASES scores have been shown not to relate to objective measures of stuttering severity such as %SS and the SSI-4.<sup>89,90</sup>

A systematic review dealing with measures of the psychological impact of stuttering on school-age children<sup>91</sup> (see Lecture Eleven) included the OASES-S. It was one of two tests with some support for its measurement properties. The authors of the review planned only to include measures of psychological impact that had their developmental data reported in peer-reviewed journals. However, they waived that criterion, and included the OASES-S in the report, even though the only supportive data for it are reported in the commercially available test manual. This is a caveat for the use of the test: its supportive data are not peer reviewed and are only available for scrutiny when the user has purchased the manual. However, another systematic review<sup>92</sup> identified eight clinical reports that presented, for 6–12-year-olds, pre-treatment and post-treatment data for the OASES with that age group. The review found four studies with “substantial treatment effects reported in the range of around 20–25% improvement post-treatment, and the one follow-up reported showing an improvement of 33% (p. 6–7).

A shortened 25-item version of the 100-item adult scale has been developed and validated for research purposes (OASES-A-R).<sup>93</sup> The authors do not recommend this scale for clinical use, stating that “the full OASES-A provides additional insights about a client’s experience of stuttering that are necessary for effective treatment planning and treatment” (p. 1).

## THE WRIGHT AND AYRE STUTTERING SELF-RATING PROFILE (WASSP)

The Wright and Ayre Stuttering Self-Rating Profile (WASSP) is another stuttering impact measure that was also designed to reflect the World Health Organization's classification system.<sup>94,95,96</sup> The WASSP is not as empirically developed as the OASES, and as yet there are no normative data. It appears to have been designed both as a clinical and research measure, with intended clinical application for assessment, and for demonstrating post-treatment client changes. The developers indicate that its contents can be used to plan treatment. Each of its 24 items is scored with a 7-point scale from *none* to *very severe*. Those items measure the domains of "stuttering behaviours (8 items), thoughts about stuttering (3 items), feelings about stuttering (5 items), avoidance due to stuttering (4 items), and disadvantage due to stuttering (4 items)" (p. 84).<sup>96</sup>

The WASSP developers report that the test has adequate reliability and validity.<sup>96</sup> It is briefer to administer than the OASES, with 10-minutes completion time reported. Purchase from the publisher is required for its use. It appears that the test is used in the United Kingdom where it was developed, but availability elsewhere currently seems to be limited.<sup>96</sup> A Turkish version<sup>97</sup> has been shown to have acceptable reliability and validity.

## SIMPLE SPEECH SATISFACTION SCALES

Often, clinical reports measure client speech satisfaction with a simple scale, such as a 9-point scale where 1 = *extremely satisfied* and 9 = *extremely unsatisfied*. Versions have been reported where 1 = *extremely happy* and 9 = *extremely unhappy*. Parents can use such scales to measure satisfaction with their children's speech. In addition to having the advantages of simplicity and validity of client self-rating scales, a speech satisfaction scale can be clinically useful because it is an overarching measure that (presumably) takes overall account of any behavioural and nonbehavioural features of stuttering that impact on the client.

A more complicated version has been suggested<sup>98</sup> where clients make a judgment about their speech using a 10-point scale where 1 = *very bad* and 10 = *excellent*, with intermediate points on the scale labelled as *bad*, *very strongly insufficient*, *strongly insufficient*, *insufficient*, *sufficient*, *more than sufficient*, *good*, and *very good*.

A simple, overarching scale<sup>89</sup> is the Satisfaction with Communication in Everyday Speaking Situations (SCESS). It involves the question "considering all the issues associated with your stuttering, how satisfied are you with your communication in everyday speaking situations at the present time?" The scale is 1 = *extremely satisfied* and 9 = *extremely dissatisfied*. The SCESS scale relates well to the OASES, to self-reported stuttering severity, but not to %SS.<sup>89</sup>

## SUMMARY

Clinical measurement is essential to assess clients and communicate with them and their parents about their stuttering. It is also essential to state treatment goals, to assess progress towards them, and to manage the maintenance of those treatment goals. Broadly speaking, clinical measurement can be categorised as "objective" or "subjective," with %SS in the former category and SR measures in the latter category. Both measures seem to capture the same dimensions of stuttering severity, with some important caveats. Stuttering severity can be measured most conveniently with SR scales. SR scales have clinical advantages related to their simplicity and validity, and their covert use with clients when needed. Speech naturalness is a useful measure for treatments that involve a trade-off between stuttering control and natural sounding speech. There are options available for measuring the impact of stuttering for clients before and after treatment. A glossary of clinical measurement procedures is presented in Appendix Two.

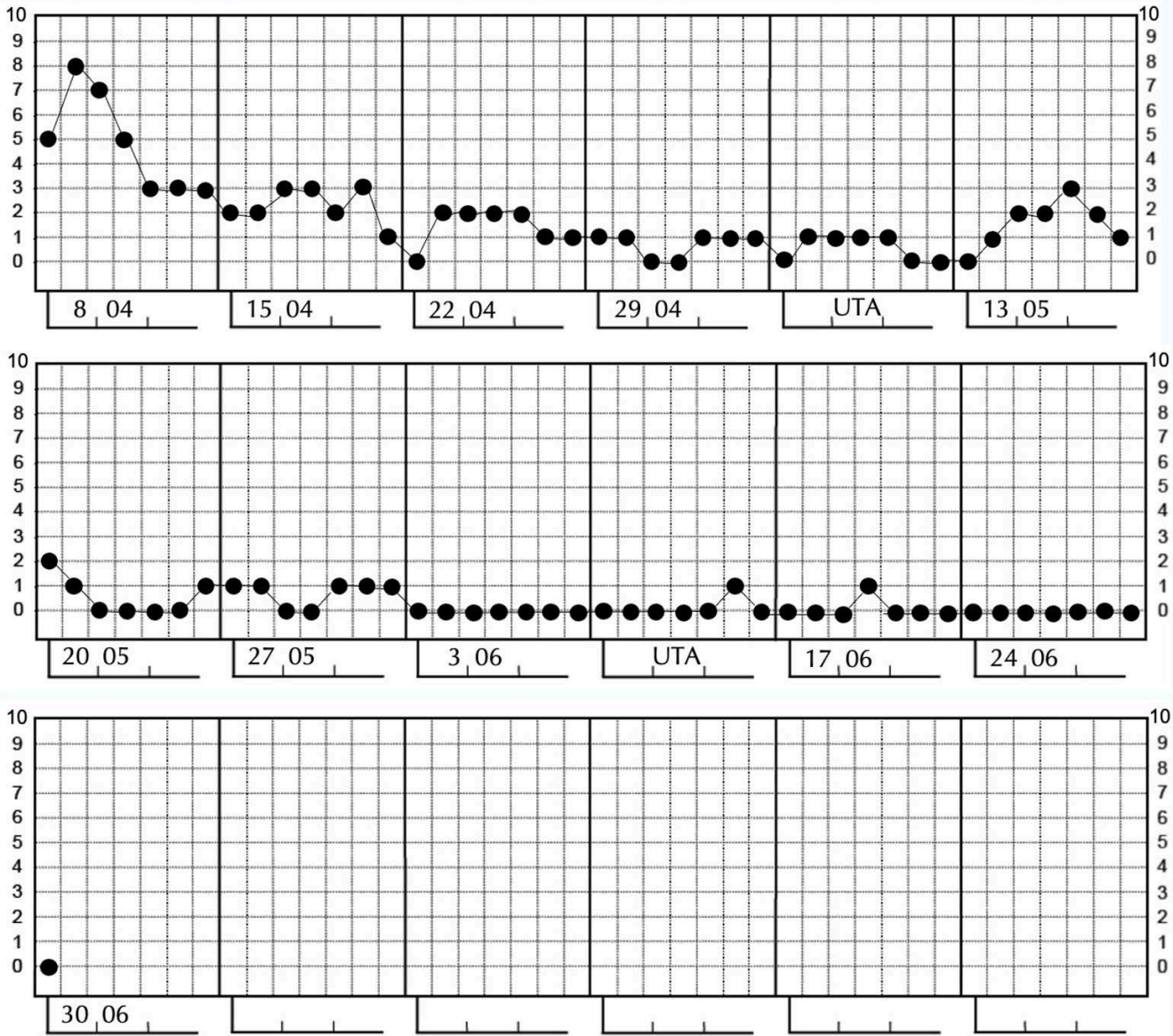
Inevitably, with advances in machine learning technology, automated measures of stuttering severity will be available for researchers, and progress is being made toward that goal.<sup>99,100</sup> It is likely that those measures will eventually be interchangeable with subjective ratings of stuttering severity. For clinicians, whether that advance will ever replace the simplicity and convenience of self-rated and

parent-rated stuttering severity, using a simple 10-point scale, remains to be seen in future decades.

**APPENDIX ONE**

SR measures during clinical management of a child with early stuttering

Closed circles are parent SR scores for each day. The numbers under the chart are the dates of the first clinic appointment of the week. UTA = unable to attend.



**APPENDIX TWO**

## Speech and impact measures for stuttering

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<i>SPEECH</i>		
Severity rating	SR	<i>A perceptual measure of stuttering severity using an ordinal scale.</i>
Percentage syllables stuttered	%SS	<i>A stutter-count measure of the proportion of spoken syllables that contains an unambiguous stuttering moment.</i>
The Stuttering Severity Instrument	SSI-4	<i>A more detailed and time consuming measure of stuttering severity than either %SS or SR.</i>
Syllables per minute	SPM	<i>A measure of speech rate.</i>
Speech naturalness	NAT	<i>A perceptual measure of how natural speech sounds using an ordinal scale.</i>
<i>IMPACT</i>		
Overall Assessment of the Speaker's Experience of Stuttering	OASES	<i>A stuttering impact measure for adults, adolescents and school-age children involving domains of general information, reactions to stuttering, communication in daily situations, and quality of life.</i>
Wright and Ayre Stuttering Self-Rating Profile	WASSP	<i>A stuttering impact measure for adults, adolescents and school-age children involving domains of stuttering behaviours, thoughts about stuttering, feelings about stuttering, avoidance due to stuttering, and disadvantages due to stuttering.</i>

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## LECTURE FIVE: EVIDENCE-BASED PRACTICE WITH STUTTERING

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### WHAT IS EVIDENCE-BASED PRACTICE?

Evidence-based practice is a health care philosophy that incorporates evidence from systematic research. Its philosophy applies not only to provision of health care to individuals who seek it, but also to government health care policy and administration.<sup>1</sup> Evidence-based practice originated with clinical medicine but has attained widespread international acceptance in many health care domains, including speech-language pathology. The best-known definition is:

Evidence-based medicine is the conscientious, explicit and judicious use of current best evidence in making decisions about the care of individual patients.  
(p. 71)<sup>2</sup>

A comprehensive video is available,<sup>3</sup> containing an interview with a speech-language pathologist, which overviews how evidence-based practice applies to health care generally.

### SPEECH-LANGUAGE PATHOLOGY AND EVIDENCE-BASED PRACTICE

Evidence-based practice has influenced the discipline of speech-language pathology. The American Speech-Language-Hearing Association is the largest professional speech-language pathology association in the world, and arguably the most influential. In 2005 it proclaimed:

It is the position of the American Speech-Language-Hearing Association that audiologists and speech-language pathologists incorporate the principles of evidence-based practice in clinical decision making to provide high quality clinical care. The term evidence-based practice refers to an approach in which current, high-quality research evidence is integrated with practitioner expertise and client preferences and values into the process of making clinical decisions.<sup>4</sup>

### THE BENEFITS OF EVIDENCE-BASED STUTTERING TREATMENT

As will be clear from the remainder of this lecture, and in fact from the entire contents of these lectures, considerable effort is needed to apply evidence-based treatment of stuttering. As stated in the preface to the lectures, they are designed to provide basic reference material for adequate health care of stuttering. As such, these lectures contain around two thousand references to scientific information, and the number is increasing with each published edition. (And that is only a fraction of relevant publications in peer-reviewed scientific journals about stuttering, which are accumulating at around 200 per year)

So, students of speech-language pathology, and junior clinicians, might well ask about what the benefits are from the effort of knowing about and accessing all that material as needed when providing health care for stuttering. Why make the effort? There are many ways to answer such a question, but the one provided here is simply that evidence-based practice links treatment and scientific research. And the benefit of that arrangement is that—to cut short a long story—scientific research improves clinical practice with stuttering, advancing it. As stated elsewhere, without that benefit, speech pathology would be “little more than an art form, handed down, essentially unchanged but for creative variations, from generation to generation of practitioners (p. 126).”<sup>5</sup> The potential reality of that projection was documented with a survey of reported treatment practices for children up to 6 years old by 121 speech-language pathologists.<sup>6</sup> The authors indicated that the reported practices did “not fulfil the ideal of evidence-based practice for stuttering managements as proposed in several international guidelines” (p. 9). Some of the questionable procedures identified were “singing,” “breathing techniques,” and “advice for sleep routines” (p. 6).

## WHAT EVIDENCE-BASED PRACTICE IS NOT

### Not a rulebook

Evidence-based practice is not a rulebook about how to provide treatment. Rather, it is a philosophy to guide treatment decisions:

Evidence based medicine is not “cookbook” medicine. Because it requires a bottom up approach that integrates the best external evidence with individual clinical expertise and patients’ choice, it cannot result in slavish, cookbook approaches to individual patient care. External clinical evidence can inform, but can never replace, individual clinical expertise, and it is this expertise that decides whether the external evidence applies to the individual patient at all and, if so, how it should be integrated into a clinical decision. (p. 72)<sup>2</sup>

### Not a source of all clinical knowledge

To fully understand how evidence-based practice influences clinical practices with stuttering, it is essential to know the limits of science. In short, systematic research is an indispensable source of knowledge to guide clinical practice, but, as stated in the previous quote, it is by no means the only source of knowledge that a clinician draws on. The abstract of Gerald Siegel’s seminal paper on the topic summarises the limits of science:

Science is a powerful tool when it addresses the kinds of questions it was designed to answer, but there are also important questions in communication disorders that fall outside the limits of science. Three such areas are discussed: Questions concerning social and personal values, questions that call for logical rather than scientific endeavors, and questions that should not be posed because we already know the answers and would not be influenced by contrary findings. (p. 306)<sup>7</sup>

Some examples of domains of clinical practice that fall outside the limits of science are empathy and emotional support for clients, listening skills, and hope and belief that treatment will help them. These are sources of knowledge cited by Siegel that relate to social and personal values and logic, and which research would not change.

### Not a replacement for common sense

A paper published in the *British Medical Journal*<sup>8</sup> makes a point about common sense during clinical practice. The authors report no evidence that parachutes improve health outcomes when jumping from aircraft, and therefore recommend that common sense might be applied to the matter of determining the health care value of parachutes.<sup>†</sup> Their point, simply, is that the quality of client care will be compromised if common sense is deleted from clinical reasoning. An example with stuttering treatment would be a client with intellectual disability. Common sense indicates that the results of treatment research might not apply to such a client as they would other clients.

## HOW TO DO EVIDENCE-BASED PRACTICE

There are many expositions about how to do evidence-based practice, but the following is a simplified summary based on the steps of the process as described at a classic source.<sup>9</sup> Evidence-based practice has been applied to health rehabilitation generally<sup>10</sup> and specifically to speech-language pathology<sup>11</sup> and stuttering treatment.<sup>12</sup> An issue of the *Journal of Fluency Disorders* was devoted to the topic of evidence-based practice and stuttering.<sup>13</sup>

### Step One: Find out what the client needs

To be fully informed, clinical judgements need to be “moderated by patient circumstances and

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<sup>†</sup> There is no mandate that scientific journals are humourless.

preferences" (p. 737).<sup>14</sup> In other words, in addition to research evidence, clinicians need to incorporate what they establish about clients' needs and their circumstances. An example of the importance of client circumstances in clinical reasoning would be a case where parents of a stuttering child are separated and share custody. In that case, evidence-based practice decisions may be different to when parents are living together, and one parent spends the day with the child during the week. Another example would be an adult who seeks control of stuttering during everyday conversations. Some clients will wish to control stuttering in certain situations only, and some clients will wish to control stuttering during more of the entire speaking day.

#### *What might clients need?*

At the most basic level, the clinician needs to determine why clients have come to the clinic. This idea of complaint-centred treatment is fundamental,<sup>15</sup> and has been applied to stuttering.<sup>16</sup> Broadly speaking, the issues that cause someone who stutters to present to a clinic will relate to either behavioural or non-behavioural matters. Either there will be some need to control stuttering, or a need to deal with a non-behavioural feature of stuttering, or a combination the two. Commonly, non-behavioural issues will involve speech-related social anxiety in some way, as will be discussed during Lectures Ten, Eleven, and Twelve.

A survey of 213 adult stutterers from around the world<sup>17</sup> provided information about this matter with a conclusion that "the vast majority consider help with speech and speech-related anxiety as an essential component of therapy" (p. 329). A survey of 502 adult stutterers<sup>18</sup> indicated that:

A majority of respondents (69.5%) indicated that they often or always have a goal of not stuttering when speaking ... A smaller percentage (36.3%) indicated that they often or always have the goal of stuttering openly and trying not to hide stuttering. (p. 329)

Additional information about this matter was presented about the clinical experiences of 71 adult members of the National Stuttering Association, which is a United States self-help group for stuttering.<sup>19</sup> Behavioural treatments to deal with stuttering were the most reported treatment received, but 49% of respondents reported receiving treatment that "involved reducing the fear of stuttering or of speaking situations" (p. 120).<sup>19</sup> Fifty-three per cent reported a combination of behavioural and non-behavioural approaches, and 25% reported non-behavioural treatment "with little emphasis on speech" (p. 120).<sup>19</sup> Thirty-three per cent "were disappointed because treatment did not address their feelings about their speech" (p. 122). Not surprisingly, the 9% who stated that "their therapist did not seek information from them in the decision-making process" (p. 122) reported dissatisfaction with the treatment process. That result was consistent with 196 survey responses<sup>20</sup> from adults, most of whom had received treatment. When asked to provide suggestions to speech-language-pathologists about desirable treatment, 17% indicated treatment geared to individual needs. Thirty percent suggested ways to control stuttering and to cope with it, and 30% advocated ways to accept stuttering and to deal with fear of it.

A report of interviews with 11 adult stutterers and 12 speech-language pathologists<sup>21</sup> explored their perceived advantages and disadvantages of changing the way of living with stuttering. Analysis of the interviews provided five overarching themes about advantages of seeking change: "enriching one's social relationships, feeling better in social interactions, developing a healthier sense of self, gaining autonomy, and communicating easier" (p. 9). Three themes about disadvantages of seeking change were "experiencing discomfort, expending resources, and recognizing that some things may not change" (p. 9). The authors argued that this information is useful for clinicians to take account of during clinical practice with adults. Interviews with 21 Kannada-speaking adults seeking stuttering treatment,<sup>22</sup> who had never received treatment, showed that all of them "expected to have some change with respect to their fluency and wanted it to be targeted in therapy" (p. 8). Around half of them "expected their affective behaviours to be more positive as the outcome of therapy" (p. 8). Affective behaviours were "anxiety, confidence, sadness, low self-esteem, negative self-evaluation, and shame" (p. 7).

A survey of 24 children and adolescents who stuttered and their parents<sup>23</sup> was conducted after

treatment. Responses were roughly evenly divided among the clients and parents about a preference “to speak without stuttering” or not. An e-Delphi Survey<sup>24</sup> of 35 adult who stuttered and 13 speech-language pathologists, who were expert in stuttering treatment, established views about core components for adult stuttering treatment. The statements where a consensus was achieved included “management of communication-related anxiety” (p. 121) and “working on speech directly to reduce the amount of stuttering” (p. 122).

With early stuttering it is usual that the prominent need will be for behavioural stuttering control. With older clients, the situation may not be as straightforward, and it might take some time to establish client need. As will be discussed during Lecture Eleven, from the school-age years through adolescence to adulthood, it seems that the likelihood of social anxiety becoming a clinically pertinent issue increase.

### **Step Two: Find the relevant evidence**

The next step is to know or find the best evidence about how to provide what clients need. There are three broad categories of such evidence that inform stuttering treatment: basic research, treatment process research, and treatment outcome research.

#### *Basic Research*

Basic research deals with the nature and the cause of stuttering. An example is research, discussed in Lecture Two, showing the possibility that a child with early stuttering will have another speech or language disorder. This information will influence assessment procedures.

Lecture Two covered epidemiological research about the nature of stuttering that clinicians may take account of when planning a treatment. For example, information about the chance of natural recovery from stuttering during the first year after onset will be a consideration in deciding when to begin early treatment, as discussed during Lecture Two and Lecture Eleven.

Another example of basic research that informs evidence-based practice is from Lecture Three, which presented information about the cause of stuttering. Research was presented that tests the veracity of various current causal explanations. In deciding whether to intervene with early childhood stuttering using a treatment based on the Demands and Capacities Model, a clinician may wish to form a view about whether that model is substantiated by basic research.

#### *Treatment process research*

Treatment process research deals with how treatment functions, as well as factors that can affect how it functions. An example from Lecture Four is that speech restructuring treatments involve a trade-off between stuttering control and natural sounding speech. This information informs clinicians when deciding whether to recommend such a treatment for a client. Another example of treatment process research that informs evidence-based practice is that percentage syllables stuttered (%SS) has been shown to be a notoriously unreliable measure, as outlined during Lecture Four. This research may prompt a clinician to reject %SS measurement during clinical practice and use severity rating methods instead.

#### *Treatment outcome research*

For stuttering, and arguably for most health care domains, clinical trials are “the most fundamental, clinically interpretable, and useful output unit of stuttering treatment research” (p. 402).<sup>25</sup> They provide research that conveys how useful treatments might be for improving the health of clients. The publication of a compelling clinical trial can change evidence-based clinical reasoning by increasing confidence in a particular treatment. Such a trial may introduce a completely new procedure to consider for clients. Or a clinical trial may show a treatment to be of limited or of no value. Clinical trials are discussed in detail shortly.

### **Step Three: Do the treatment and evaluate its effects**

The final step in the evidence-based reasoning process is to administer a treatment and evaluate its



effects. This can be done with regular application of clinical measures outlined during Lecture Four. The graph in Appendix One of that lecture is an example of clinical measurement used to establish that a child responded favourably to treatment and continued to do so. If these clinical measures had not shown that a change was apparent after several weeks of treatment, the clinician would have needed to problem-solve and make changes to the treatment process.

## SCIENTIFIC STANDARDS FOR CLINICAL EVIDENCE

### Peer-reviewed scientific journals

Clinician consumers of research need to determine whether research is sufficiently credible to warrant consideration. A rule of thumb here is whether the research has been published in a peer-reviewed journal that is listed in a credible data base such as PubMed or the Web of Science. Publication sources apart from peer-reviewed journals are sometimes referred to as the “grey literature,” and include student theses, books, book chapters, and internal institutional reports.

That said, the standards of scientific journals, and the rigour of their peer review, are not at all uniform. For example, some peer-reviewed journals do not meet standards for inclusion set by prestigious databases such as the Web of Science.<sup>26</sup> Certainly, published articles should not be accepted at face value; many of them should be disregarded. In any event, there is cause to regard with serious reservation any research that has not been reviewed and endorsed by peers within the scientific community and cleared for publication by the editor of a respected journal. Recent years have seen the emergence of harshly but appropriately named “junk” research in “junk” journals.<sup>27</sup>

### Hierarchies of evidence

After a decision about whether research is sufficiently credible to warrant consideration, clinician consumers of research then need to make a further judgement about the standard of the research. To inform that decision, there are some generally accepted overarching standards for health care research, presented as hierarchies of evidence. That said, the earlier point needs reiteration, that evidence-based practice is not a rulebook for decision-making. A judgment about the quality of published clinical research will vary from clinician to clinician.

Prominent examples of evidence hierarchies are the University of Oxford Centre for Evidence-Based Medicine,<sup>28</sup> the National Health and Medical Research Council of the Australian Government,<sup>29</sup> and the Cochrane Consumer Network.<sup>30</sup> A common theme appearing in all those is that at the top of the hierarchy—the most convincing evidence for health care—is a systematic review that synthesises evidence from numerous randomised controlled trials using meta-analysis. Scientific journals and textbooks regularly publish systematic reviews, and The Cochrane Collaboration<sup>31</sup> is a well-established and trusted online source of systematic reviews.

Apart from a systematic review, those hierarchical classifications<sup>28,29,30</sup> then specify that a minimum of one randomised controlled trial is the next best level of evidence. They specify methodological variants of randomised designs as less compelling, such as pseudo-randomisation and cluster randomisation. Non-randomised designs are relegated to lower levels. These include case control and cohort studies, followed by case studies of groups and individuals and time series studies of individuals. The Oxford Centre for Evidence-Based Medicine<sup>28</sup> specifically places “expert opinion” as the least admissible source of evidence for the value of a treatment. In the context of stuttering treatment, reliance on expert opinion rather than on scientific research to guide treatment has been described as assertion-based practice.<sup>32</sup> Experts, many of them charismatic, commonly proclaim the merits of certain treatments when there is no credible research evidence to support their claims. The media often endorses such claims because of their charismatic nature.

### Critiquing a research paper

When a clinician decides that a published research paper is worth considering, it is then necessary to make a value judgement about its methodological credentials. This requires a basic understanding of

research methods, presumably attained during university professional preparation. For papers with various research designs, checklists are publicly available.<sup>33</sup> There is a website available that provides methodological critiques of treatment reports in speech-language pathology,<sup>34</sup> and gives each report a quality rating. The site was modelled on similar sites for clinical psychology and physiotherapy. It includes critiques of many papers dealing with stuttering.

For most common health problems, there are specifically designed standards for health care research. In the case of stuttering treatment research, a detailed, 136-item checklist has been proposed<sup>35</sup> which clinicians can use to critically appraise stuttering treatment research evidence. The authors reported that inexperienced judges can use it reliably. The checklist was based on research standards that have historically been endorsed by leading scholars and researchers in the field.

## CLINICAL TRIALS OF STUTTERING TREATMENT

### What is a clinical trial?

Because of the prominence of the clinical trial in health research, it is useful for clinicians to have some criteria for determining what is and what is not a clinical trial. There are many definitions available. The World Health Organization defines a clinical trial as

... any research study that prospectively assigns human participants or groups of humans to one or more health-related treatments to evaluate the effects on health outcomes.<sup>36</sup>

The National Institutes of Health, which funds health research in the United States, defines a clinical trial this way:

A prospective biomedical or behavioral research study of human subjects that is designed to answer specific questions about biomedical or behavioral treatments (drugs, treatments, devices, or new ways of using known drugs, treatments, or devices). Clinical trials are used to determine whether new biomedical or behavioural treatments are safe, efficacious, and effective.<sup>37</sup>

The major Australian government health funding bodies and Australian Universities involved with health research define it this way:

A clinical trial is a form of human research designed to find out the effects of a treatment, including a treatment or diagnostic procedure. A clinical trial can involve testing a drug, a surgical procedure, other therapeutic procedures and devices, a preventive procedure, or a diagnostic device or procedure. (p. 33)<sup>38</sup>

### Definition of a clinical trial for these lectures

For the purposes of these lectures, the following definition of a clinical trial is used because it is designed specifically with reference to stuttering treatment, and it incorporates reasonable consensus from within the speech-language pathology discipline:

A clinical trial of a stuttering treatment is (a) a prospective attempt to determine the outcome or outcomes of (b) at least one entire treatment with (c) at least one pre-treatment and one follow-up outcome of at least 3 months in the case of a reported positive outcome, and (d) where outcomes involve speech observations that are independent of treatment and derived from recordings of conversational speech beyond the clinic. (p. 404)<sup>25</sup>

This definition contains an essential component of speech measurement outside the clinical setting. At present the gold standard for doing so involves the %SS measure, as outlined during the previous lecture. However, two recent reports introduced some potential flexibility here with findings that, for early and persistent stuttering, %SS and severity rating scores beyond the clinic do equally well for documenting the results of stuttering treatment during clinical trials.<sup>39,40</sup> The caution here, though, is that these findings have yet to be replicated.

Although this definition of a clinical trial is used throughout these lectures, it is an arbitrary perspective

on the matter. The many reviews of the evidence for stuttering treatment efficacy present differing views about what should be regarded as a clinical trial of stuttering treatment. For example, this report<sup>41</sup> presents a far more liberal view of the matter than that just described, including reports that contain “any outcome relating to a positive effect on ... communication or ... social and emotional wellbeing” (p. 678)<sup>41</sup> without regard to follow-up or whether speech measures were collected beyond the clinic. At the other extreme is the 136-item checklist mentioned previously.<sup>35</sup> A middle ground position is a 29-item checklist presented by other authors.<sup>42</sup>

That said, some details about the presently used working definition<sup>25</sup> are as follows.

### **Clinical trial standards**

#### *Prospective methods*

Using the definition just outlined,<sup>25</sup> or in fact any definition, a retrospective study would not be regarded as a clinical trial of a stuttering treatment. Examples of retrospective studies would be file audits of previously treated clients,<sup>43</sup> and clinical follow-up of such cases.<sup>44</sup> That is not to say that retrospective reports of stuttering treatment outcome are not useful publications to consider during evidence-based practice. To the contrary, they are useful demonstrations of the potential efficacy of a treatment and may be important preliminary precursors to a clinical trial and may in some circumstances be considered during evidence-based clinical reasoning.

#### *Study of complete treatments*

Many reports of stuttering treatment do not report about the entire treatment. Some such reports are more appropriately termed clinical experiments than clinical trials. Again, that is not to say that clinical experiments are of no value during evidence-based practice. For example, a clinical experiment may produce evidence that a treatment can stop the developmental course of stuttering.<sup>45</sup> However, clinical experiments do not contribute information about the outcome of the entire treatment.

#### *Beyond-clinic speech measures*

If clinical behaviour change is part of treatment, it needs to be measured beyond the clinic to be sure that it has really occurred.<sup>46</sup> This is because of what is known as discriminated learning, which refers to the learning of behaviour change that occurs in the clinical setting where it is taught, but not necessarily outside the clinic. Regardless, it is common sense that clinical trials need to show intended stuttering reductions outside the clinic, because treatment needs to improve speech during everyday life. Authorities in the field of stuttering agree about this matter.<sup>47</sup> The most common behavioural speech measure for clinical trials of stuttering treatment is blinded %SS scores.

#### *Follow-up period*

The clinical trial definition presented earlier specifies that speech measures are collected after a follow-up period of at least 3 months. That is a liberal requirement. Normally, for a treatment to be regarded as useful, researchers would need to demonstrate that clinically significant treatment effects remain in place for a year or more after treatment. Often, clinical trials of stuttering treatment do report data with such follow-up periods.

## **PHASES OF CLINICAL TRIAL DEVELOPMENT**

### **Phases I to IV**

Clinical trials normally proceed with four developmental stages, from Phase I to Phase IV. The components of each stage, particularly the number of participants involved, differs from discipline to discipline. For example, there are normally more participants in clinical trials of drugs than in clinical trials of stuttering treatments. There is a short and readable introduction to clinical trials that was written specifically for a speech-language pathology audience.<sup>48</sup> It is a general rule that clinicians can have more confidence in the results of clinical trials when they are at a more advanced phase of

development. Given equivalent methodological rigour, a Phase III clinical trial is far more compelling than a Phase II clinical trial.

### The CONSORT Statement

Standards for an acceptable clinical trial are specified in the Consolidated Standards of Reporting Trials (CONSORT) Statement.<sup>49</sup> The group who drives and maintains the CONSORT Statement comprises scientists and medical journal editors. Many prestigious medical journals will not accept a clinical trial unless it conforms to the CONSORT Statement, and there is an increasing trend for authors of clinical trials, including trials of stuttering treatment, to indicate that their trial design conforms to those guidelines.

### Phase I clinical trials

#### *An early stage of treatment development*

Phase I clinical trials are normally the first stage in a sequence of treatment development. For stuttering research, they normally involve only a few participants. Their purpose is to develop preliminary evidence that justifies continued development of the new treatment. The kinds of information sought with Phase I trials are whether clients will comply with the treatment, whether it is safe, and whether there is any suggestion that there might be a treatment effect.

#### *Non-randomised*

Phase I trials are not randomised, meaning that there is only one group who receives the treatment. Measures are made pre-treatment and then post-treatment. Because of the few participants who are involved in Phase I clinical trials, and because they are non-randomised, they are the least compelling of clinical trial evidence.

### Phase II clinical trials

#### *A “green light” for further trials*

The next stage of clinical trial development is the Phase II trial. These normally have more participants than Phase I trials and are designed to collect more convincing evidence of any potential treatment effect. A Phase II trial can give a “green light” for the conduct of a Phase III clinical trial. During Phase II trials, the safety and viability of the new treatment continue to be monitored, the treatment is adjusted according to need, and the final treatment protocol is developed.

#### *Can be randomised*

Phase II trials are normally non-randomised, but they can be randomised.<sup>50,51</sup> Randomisation means that there are two groups, often a control group who receives no treatment, and an experimental group who receives the treatment being developed. A variation is for two or more treatments to be compared against each other, possibly with that comparison involving a no-treatment control group. Randomisation of Phase II trials is a method with controversial features (Chapter 13).<sup>52</sup>

#### *The importance of randomisation*

There is a well-known effect where non-randomised trials overestimate the true effect size.<sup>53</sup> In other words, they suggest that the treatment is better than it really is. The most common sources of bias in non-randomised trials are placebo effects and regression to the mean. Regression to the mean is where those who stutter seek clinical help when their stuttering is at its worst, only to improve subsequently because of natural variation. For clinical trials involving early childhood stuttering, there is another ever-present source of bias: children in such trials might recover from stuttering not because of the treatment but because of natural recovery (see Lecture Two).

Randomisation ensures that the trial is as free of bias as is possible. However, clinical trials can never be completely free of bias. This is because participants in a clinical trial need to volunteer to be involved in the research, and such volunteers may be unlike those who generally present to clinics for speech treatment.

An advantage of a randomised Phase II trial is that it enables a mathematical calculation of what the true effect size is, in ways to be discussed shortly. When beginning a Phase III trial, it is necessary to have some idea of the effect size because it is used mathematically to determine how many participants are needed for such a trial.

### Phase III clinical trials

#### *The “gold standard”*

Phase III clinical trials are often referred to as the gold standard of clinical trial research. They are expensive and logistically taxing for researchers to undertake. A comprehensive guide to Phase III clinical trials of stuttering, written specifically for a speech-language pathology audience, is available.<sup>54</sup> Salient points from this guide are summarised below.

#### *Recruitment*

First, participants are recruited to the trial and give their consent to be randomised to one of the groups. For most trials of stuttering treatment, participants are recruited from clients presenting to a clinic, although newspaper and other types of advertising can publicise the trial. After recruitment, the pre-randomisation measure or measures are collected. These are referred to as pre-randomisation measures, not pre-treatment measures, because in a randomised controlled trial involving a no-treatment group, half the participants in fact receive no treatment.

#### *Randomisation*

Next, an independent person randomises the participants. Ideally, the independent person is a biostatistician, or a researcher who implements a randomisation method that has been prescribed by a biostatistician. There are several different ways of randomising participants to trials, according to features of the trial design and how many participants there are.

#### *Treatment arms*

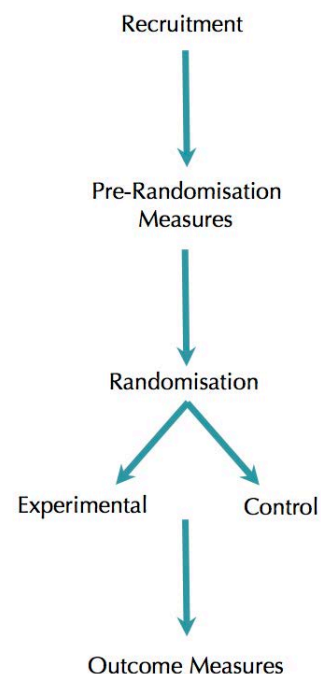
As with Phase II trials, there can be three or even more groups, or *arms* to use the correct term. Treatments can be compared to each other or to a control arm. A trial with a no-treatment control arm and an experimental treatment arm compares the experimental treatment with no treatment. A trial can compare two treatments that are completely different, or it can compare variations of the same treatment. An example of the latter would be a clinical trial comparing a treatment given to participants in a clinic to a treatment given to participants by video telehealth. With clinical trials that compare two treatments there also can be a no-treatment control arm.

#### *Primary outcomes*

In a clinical trial, outcome measures are fundamental to how the outcome of the trial is judged. The researchers who do the trial consider the primary outcome to be the most clinically important among several outcomes that can be measured. The CONSORT Statement strongly suggests that a randomised trial should have no more than one primary outcome. The reason given for this is that, from mathematical and logical viewpoints, more than two primary outcomes makes it difficult to interpret the results of the trial.

#### *Secondary outcomes*

Although ideally there should be one primary outcome for a randomised trial, there can be several secondary outcomes. Secondary outcomes are measures that are used to complement the primary outcome as measures of interest. For example, a randomised trial of a treatment to control stuttering may have %SS as the primary outcome, and secondary outcomes might be clinician and client SR



scores, along with NAT and SPM scores (see Lecture Four).

### *Effect sizes*

The most trustworthy estimate of effect size is obtained from a randomised trial where the response of one or more treatment groups is compared with the response of a no-treatment control group. Effect sizes can be estimated mathematically. A basic method for estimating effect size is Cohen's  $d$ .<sup>55</sup> This is the difference between the mean primary outcome of the experimental and control group divided by the average standard deviation of the two groups. This gives a measure of effect size in standard deviations. By convention, a Cohen effect size of 0.2 is regarded as small, 0.5 medium, and 0.8 and greater as large. Cohen effect sizes can be larger than 1.0. There are several variations of the method for calculating Cohen's  $d$ .

Another way to measure effect size is the *odds ratio*, and this has been reported in several trials of stuttering treatment. This is a measure of the odds having a certain health outcome. For example, an odds ratio of 6.5 in a randomised controlled trial might mean that the group who received the treatment had 6.5 times greater odds of attaining below 1.0 %SS at post-treatment than the group who received no treatment. An odds ratio of 4.0 is generally considered to be favourable. There are related indices of effect size that have not yet appeared in the stuttering treatment literature: absolute risk reduction, relative risk, relative risk reduction and "number needed to treat."

### *Three ways effects can be significant*

Measures of effect size are normally accompanied by a measure of statistical significance. For example, a report might indicate that a treatment group had better odds of attaining below 1.0 %SS than the control group, and report that  $OR=7.5$ ,  $95\% CI=4.7-10.9$ ,  $p<.0001$ . The way to read this is that the odds ratio was 7.5 with a 95% confidence interval of 4.7–10.9. In other words, the plausible range for the true odds ratio value, with 95% certainty, was somewhere between 4.7 and 10.9. If the confidence interval contains zero, there is no evidence of a difference between the groups.

However, this is not the entire story about the significance of effect sizes for stuttering treatment, or any treatment.<sup>56,57</sup> A difference may be statistically significant but of no practical significance. For example, a group may have a mean pre-treatment score of 12.7 %SS and a post-treatment score of 10.3 %SS. This could well be a statistically significant difference; however, such a small change is unlikely to be of any clinical significance. The term *personal significance*<sup>57</sup> takes account of the extent to which—regardless of numbers—a treatment remedies the life issues and consequent presenting clinical complaints that it causes.

In clinical psychology, the *reliable change index* is commonly used to define what is a clinically significant change,<sup>58</sup> using a statistical method to determine whether a pre-treatment to post-treatment change is statistically believable. The procedure requires information about the standard error, which is the standard deviation of a sampling mean. Such data are available for stuttering<sup>59</sup> and, hence, it is possible to use this procedure for %SS data before and after treatment, and this method has been demonstrated with stuttering treatment.<sup>60</sup> The reliable change index also allows assessment of whether treatment moves the client from a dysfunctional range to a functional range of performance on a clinical measure. The authors of the previous paper<sup>60</sup> argue—contentiously—that this can be done for %SS scores by using 0.5 %SS as the cut-off score for typical speech after treatment.

### *Drop-outs*

With randomised controlled trials, researchers are required by the CONSORT statement to report drop-outs. Knowing how many participants dropped out of a trial, and from what arm, influences the confidence that can be placed in the results. If, for example, one quarter of all participants in the experimental arm dropped out of treatment, that would need to be considered when evaluating the results of the trial.

One approach to dealing with clinical trial dropouts is by *intention-to-treat analysis*.<sup>61</sup> This means analysing trial outcomes of participants according to the treatment group into which they were

randomised, regardless of whether they completed treatment or what treatment they completed.<sup>†</sup> One way to do this is by applying *last observation carried forward*. This means that if a participant drops out after collection of pre-randomisation speech measures, for example, then those pre-randomisation speech measures are included as post-randomisation measures. This provides a conservative rather than a liberal estimate of effect size.

### Phase IV clinical trials

Once Phase III clinical trials have established the value of a particular treatment, Phase IV clinical trials may be used to determine how well they work among the community of clinicians who need to use them during everyday professional practice. Phase IV of clinical trials development is often referred to as translational research.

When discussing the merits of treatments, the terms *efficacy* and *effectiveness* are often used interchangeably. However, strictly speaking, they mean different things. Efficacy refers to a demonstration of the effects of a treatment under the specialised conditions of a clinical trial as conducted by professional researchers. Usually, clinical trials are conducted in dedicated research facilities, with specially trained clinicians, and explicit attempts are made to ensure that the treatment is being done correctly. The strictly correct use of the term effectiveness refers to whether a treatment is useful when used by a community of professional clinicians who operate in the “the real world” of treatment, as demonstrated by Phase IV clinical trials.

### KEEPING UP TO DATE WITH STUTTERING RESEARCH

A recent discussion<sup>62</sup> concluded that—for generalist clinicians at least— it is challenging to keep up with stuttering research as it is being published. This is because generalist speech-language pathologists provide treatment services not only for stuttering but for a range of speech and language disorders, of many kinds, for several ages. Consequently, the discussants offered the following recommendations about how this task might be achieved in the most time efficient manner. The following recommendations in that paper (p. 8) focus on treatment efficacy and clinical trials, and five categories of basic research: quality of life, psychological impact, epidemiology, stuttering causality, bilingualism and multilingualism.



- Obtain and read the stuttering management guidelines for the professional speech-language pathology association in your country and check regularly for updates.
- Obtain and read *reviews and systematic reviews* of stuttering treatment efficacy that are pertinent to your caseload (child, adolescent, adult). These can be obtained with searches of speech-language pathology databases.<sup>63,64</sup> To ensure comprehensiveness, additional searches of Google Scholar and PubMed are needed. (search terms “stutter\*” and “stammer\*” and “review” and “treatment” and “child” or “adolescent” or “adult”).
- Obtain and read *papers* that are pertinent to your caseload that report *clinical trials*, using a liberal definition of a clinical trial. A clinical trial is defined as a prospective evaluation of an entire treatment, with beyond-clinic outcomes and follow-up at least 3 months.<sup>25</sup> Search Google Scholar and PubMed (stutter\*” and “stammer\*” and “review” and “trial
- Evaluate the quality of the *clinical trials* above. Use these publicly available checklists<sup>33</sup> for randomised controlled trials, quasi-experimental studies, case reports, systematic reviews.

<sup>†</sup> Sometimes participants “drop-in,” which means they receive the experimental treatment even though they were not randomised to receive it.

- Selecting from abstracts of *papers* about stuttering, obtain and read those papers pertinent to your caseload that report *case studies, file audits, and expert opinion about stuttering treatment*. Abstracts of all papers about stuttering can be obtained with Google Scholar and PubMed data base searches: (“stutter\*” and “stammer\*”).
- Regardless of your caseload, obtain and read *reviews and systematic reviews* about the following basic science domains pertinent to evidence based practice: *quality of life, psychological impact, epidemiology, stuttering causality, bilingualism and multilingualism*. Search Google Scholar and PubMed (stutter\*” and “stammer\*” and “review” and “quality of life” and “psychology\*” or “epidemiology” or “cause” or “theory” or “bilingual\*” or “multilingual\*”).
- Obtain and read papers identified from these basic science domains pertinent to clinical practice: *quality of life, psychological impact, epidemiology, stuttering causality, bilingualism and multilingualism*. Search Google Scholar and PubMed as above.
- With a colleague, mentor, or stuttering interest group, online or in person, discuss recently emerging material from the sources above.
- Search the archives of podcasts for information about these topics, and listen to them: *clinical trials, treatment efficacy quality of life, psychological impact, epidemiology, stuttering causality, bilingualism and multilingualism*.
- Attend as many as possible online or in-venue conferences and training events that deal exclusively with stuttering or which contain substantial components about stuttering.

## SUMMARY

Evidence-based treatment practices are an ethical requirement of the speech-language pathology discipline. Evidence-based practice incorporates judgements about the best research evidence to inform clinical management decisions with clients. Clinician judgements about the quality of research evidence can be informed by publications in peer-reviewed scientific journals, accepted hierarchies of evidence strength, and detailed methodological critiques of research publications. Clinical trials are the fundamental output of clinical research that informs treatment practices. Clinical trials evolve treatment development in four stages that move from preliminary evidence of treatment effects to evidence of population effectiveness. Reviewing the scientific literature requires a time commitment by clinicians, but that time commitment is worthwhile in terms of its rewards.



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## LECTURE SIX: EVIDENCE-BASED INTERVENTIONS FOR EARLY CHILDHOOD STUTTERING

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### EARLY INTERVENTION WITH TELEHEALTH

#### Telehealth

Current early childhood stuttering interventions were developed for the traditional format of weekly clinic visits. The term *telehealth* refers to treating clients when they are not in the clinic. There are reviews available of telehealth in speech-language pathology with specific reference to stuttering treatment.<sup>1,2</sup> Professional speech-language pathology associations have also reviewed telehealth service provision and associated professional issues with it.<sup>3,4,5</sup>

During the Covid pandemic, 106 United States and Canadian speech-language pathologists<sup>6</sup> were surveyed about delivering the Lidcombe Program (a treatment to be discussed shortly) with telehealth. A majority of 94% indicated that they would include telehealth in future Lidcombe Program delivery. They reported the treatment to be easily adaptable to telehealth, with benefits including “time efficiency, flexibility of scheduling, and improved clinical processes” (p. 1). Reported challenges involved technology, establishing a clinical relationship, and identifying mild stuttering.



Telehealth seems viable for presenting the Lidcombe Program in a school setting. According to interviews of five school-based Australian clinicians,<sup>7</sup> the key issues were (1) understanding and managing the required technology, (2) logistics of the procedure in a school setting, (3) support from colleagues and the school, and (4) establishing family engagement with the telehealth model.

Video telehealth is currently accessible using laptops, tablets, and smartphones. The number of households with internet connections is increasing rapidly, and internet transmission rates are constantly improving.

#### Advantages of telehealth for early childhood stuttering intervention

##### *Limited infrastructure needed*

The traditional clinical infrastructure for pre-school children and parents is not needed for telehealth. All that is needed is a workspace, an internet connection, and a screen. Software that the clinician can use to video record entire treatment sessions is either free of charge or inexpensive.

##### *Benefits of home clinician contact*

Parents do not need to prepare their child for travel to the clinic, and either arrange childcare for siblings or bring them along to the clinic. There is evidence that parents can find this to be a significant burden added to doing the treatment in a clinic.<sup>8</sup> Additionally, a significant clinical advantage of telehealth is that the clinician sees the parents doing treatment with their children in their own homes. This makes clinical sense, not only from the perspective of common sense, but also in terms of generalisation theory,<sup>9</sup>

##### *Families isolated from in-clinic services*

In large and sparsely populated countries, there will be many families who are isolated from in-clinic treatment services. Telehealth is useful for the many families who are in such a situation. In Australia,

for example, one third of families live rurally, and, apart from geographical isolation, such isolation presents many barriers to treatment access.<sup>10</sup>

#### *Facilitates specialisation*

Telehealth facilitates clinical specialisation. It enables a clinician with specialised skills for stuttering to treat children who are located anywhere in the country. Additionally, with flexible working hours, a clinician can treat children anywhere in the world. Telehealth can therefore facilitate a clinician becoming known nationally and internationally for specialised stuttering treatment services.

#### *Telehealth and stepped care*

The advantages of telehealth with early stuttering intervention suggest that it may have a place within a stepped care model of healthcare delivery. That model contains two fundamentals.<sup>11</sup> First, it provides the simplest and most cost-efficient method of health care that is efficacious. Second, it is self-correcting so that clients progressively escalate to more resource intensive, and more costly, models of health care if they are shown to need it. So, if families do not respond to telehealth early stuttering intervention, they might then go to a clinic each week. Or an intervening step might be that telehealth intervention is supplemented by occasional clinic visits. Treatment can begin at any step, not necessarily the first. Work has begun to develop a standalone internet Lidcombe Program treatment that does not require a clinician,<sup>12</sup> suggesting the possibility of such treatment as the first intervention in stepped care. The stepped care intervention model has been shown efficacious with management of several conditions,<sup>13,14,15,16,17</sup> but there seems to have been only one description of the stepped care concept applied to stuttering.<sup>18</sup>

### **THREE EARLY CHILDHOOD STUTTERING TREATMENTS THAT ARE SUPPORTED BY CLINICAL TRIALS**

Three early childhood stuttering interventions that have clinical trial evidence to support them, using the definition of a clinical trial presented in Lecture Five,<sup>19</sup> are reviewed here. The treatments are supported by either Phase I, Phase II or Phase III evidence. These treatments are the Lidcombe Program, the Westmead Program, and two treatments that are conceptually similar to each other: Palin Parent-Child Interaction Therapy and RESTART-DCM treatment. The relative strengths and limitations of the three treatments are discussed, expanding on a previous discussion of the matter.<sup>20</sup> In the next lecture, the clinical research that supports the three treatments will be presented. Of all treatments considered, the Lidcombe Program has the most extensive evidence base of clinical research, so it is considered with the most detail.

### **THE LIDCOMBE PROGRAM**

#### **Background**

##### *Basic research that led to its development*

Lecture One described an extensive body of basic stuttering research from the 1950s and 1960s showing that response contingent stimulation can reduce stuttering to a clinically useful extent. During the 1970s, laboratory experiments showed that verbal response contingent stimulation could be used with children and could obtain similarly useful stuttering reductions.

The most famous of these experiments was the so-called puppet study of 1972,<sup>21</sup> which adapted a technique developed a decade earlier.<sup>22</sup> The researchers set up an illuminated puppet that conversed with child stutterers. Under the experimental conditions the light was turned off during moments of stuttering, effectively making the puppet disappear. The researchers showed that this ingenious application of verbal response contingent stimulation successfully controlled the early childhood stuttering of two children. That control generalised beyond the laboratory and was maintained for around 1 year. The Lidcombe Program involves the operant method of parents providing verbal response contingent stimulation to children who stutter (see Lecture One). An early description of this

type of approach appeared in 1971.<sup>23</sup>

#### *Clinical resource materials*

These are located at the website of the Australian Stuttering Research Centre.<sup>24</sup> The *Lidcombe Program Treatment Guide* is a clinical reference for the treatment, and the *Lidcombe Program Brochure* describes the treatment. There is a clinical severity rating (SR) chart for parents and clinicians: *Child Stuttering Severity Chart eForm*. That form is reproduced in Appendix One of this lecture. The website also contains the *Stuttering Treatment Activity Guide*, and the *Stuttering Treatment Activity Guide-For School-age Children*, and *Considerations for Problem Solving in the Lidcombe Program*. These materials have been translated into several languages.

Information about the Lidcombe Program has been made available beyond the speech-language pathology discipline to general and paediatric medical practitioners with overviews in medical journals.<sup>25,26,27</sup> Clinical checklists are available for clinicians to use to ensure they are doing the treatment correctly. One of these was validated by users,<sup>28</sup> and another is presented with case studies of its use.<sup>29</sup> One of those checklists<sup>28</sup> contains 63 items and is reproduced at the end of the *Lidcombe Program Treatment Guide*.

The *Lidcombe Program Trainers Consortium*<sup>30</sup> has members in 12 countries and provides postgraduate training for the treatment. This training involves two days of instruction and demonstration, often with subsequent clinical follow-up. The developers of the Lidcombe recommend that clinicians do not attempt it without Consortium training. The Lidcombe Program is endorsed by the professional associations of English-speaking<sup>31,32,33</sup> and non-English speaking countries.<sup>34,35,36,37,38</sup>

## Overview

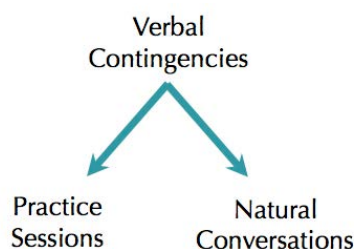
### *A behavioural treatment*

The Lidcombe Program is a behavioural treatment, designed to deal with children's stuttered speech. It uses operant methods, even though, as discussed during Lecture One, stuttering is not freely emitted problem behaviour and in no proper sense is it an operant.

The Lidcombe Program is unlike the other two treatments considered during this lecture. It does not require children to change their customary speech pattern in any way, and it does not require any change to the customary living environments of children to remove features of those environments thought to cause or sustain stuttering.

### *Parents give verbal response contingent stimulation*

Parents do the Lidcombe Program with training and supervision by a clinician. It involves parents giving their children verbal response contingent stimulation—verbal contingencies—for not stuttering and for stuttering. They do this during practice sessions with their children, designed specifically for this purpose, and during naturally occurring conversations with their children. On most occasions it is the parents who give the treatment to their children, but sometimes it may be caregivers.



### *Clinical measurement*

Regular measurement of children's stuttering severity occurs during the Lidcombe Program.

*Parents have an appointment each week*

During each weekly consultation the clinician teaches parents how to do the treatment and ensures that it is being done properly. The treatment guide<sup>24</sup> specifies what occurs during each clinic consultation, and in what order.

*Treatment goals during Stage 1 and Stage 2*

Lidcombe Program treatment goals are no stuttering or nearly no stuttering for a long time. The goal of Stage 1 is no stuttering or nearly no stuttering, and the goal of Stage 2 is for that to be sustained for a long time. Stage 2 of the treatment is sometimes referred to as maintenance.

**The severity rating (SR) scale<sup>†</sup>***Treatment goals specified with SR scores*

Parents give their children a SR for each day and clinicians give a SR during each clinic appointment. They use the SR scale described during Lecture Four: 0 = *no stuttering*, 1 = *extremely mild stuttering*, 10 = *extremely severe stuttering*. Lidcombe Program treatment goals are specified with SR scores.

To progress to Stage 2, the following two criteria need to be met for three consecutive clinic appointments that are 1 week apart: (1) parent SRs of 0–1 during the week preceding the clinic appointment, with at least four of those seven SRs being 0 and (2) clinician SRs of 0–1 during the clinic appointment. A minimal requirement during Stage 2 is for parents to record SRs only during the week preceding the clinic appointment. However, the clinician may direct parents to record SRs more often during Stage 2. (p. 10)<sup>24</sup>

*A flexible measurement*

Parents give a SR to their children's speech for each day. This measurement procedure has some flexibility, as outlined in the treatment guide:

Variations of the SR procedure can be added to the treatment process if the clinician thinks it would be useful, commonly one SR for the morning and one for the afternoon. Clinicians may wish parents to use supplementary SRs for a particular speaking situation that occurs each day, such as at dinner and bath time, and shopping. These are recorded in addition to the daily SRs. Other options are for parents to record a highest and lowest SR for each day. (p. 2)<sup>24</sup>

*SR scores to determine if treatment is working as planned*

Severity rating scores are used to check that children's clinical progress is satisfactory. If progress is not satisfactory then SR scores will alert the clinician, and the problem can be solved. Such problem solving is a routine part of Lidcombe Program administration.

*Accurate parent severity ratings are essential*

It is essential for parents to use the SR scale accurately, or the treatment cannot work properly. If parents underestimate their children's stuttering severity with the scale, it can result in them being admitted to Stage 2 prematurely, before they have attained the SR treatment goals just outlined. For example, parents might give an average SR of 0.3 for the week before a clinic appointment when the appropriate average SR is 2.3. The opposite situation, where parent SRs are too high, would waste clinical resources by causing the child to take longer for treatment than necessary.

*Parent severity rating training*

It is a simple matter to prevent problems where parents use the SR scale inaccurately. To quote from the treatment guide,

During the first clinic appointment, after the clinician has explained the SR scale, the parent or the clinician, or both, converse with the child for a few minutes

<sup>†</sup> Prior to 2015 the Lidcombe Program used a 1–10 SR scale, and publications before then contain that version of it.



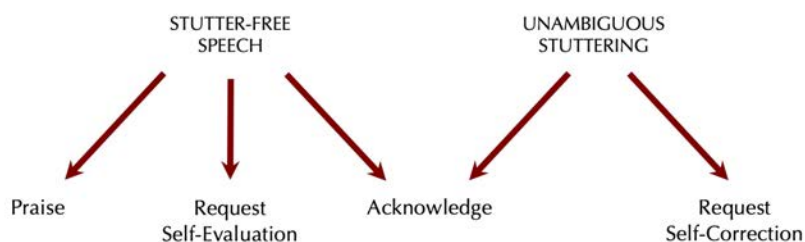
until the child displays a reasonably representative amount of stuttering. After a few minutes the clinician asks the parent to assign a SR to the speech sample. The clinician indicates whether that is an appropriate score and, if necessary, suggests a different score. All subsequent clinic appointments begin with the parent conversing with the child, the parent assigning a SR score, and the clinician either confirming that the score is appropriate or providing corrective feedback. (p. 3)<sup>24</sup>

### Percentage syllables stuttered (%SS)

There has been a recent recommendation, with theoretical and empirical justification, for why %SS is no longer an essential part of the Lidcombe Program.<sup>39</sup> However, for reasons outlined in that publication, some clinicians may prefer to use %SS during each clinic session when using the treatment.

### Parent verbal contingencies

There are five verbal contingencies in the Lidcombe Program that the treatment guide specifies as essential. Three of these are verbal contingencies for stutter-free speech, and two are for unambiguous stuttering moments. An overview of the essential Lidcombe Program verbal contingencies is shown in the figure below.



#### *Verbal contingencies for stutter-free speech*

Verbal contingencies for stutter-free speech are central to the Lidcombe Program because, above all else, children must enjoy the treatment for it to work properly. Parent verbal contingencies for stutter-free speech are inherently positive and supportive, which is essential for clinical dealings with children who have early stuttering.

The first parent verbal contingency for stutter-free speech is *praise*. The clinician teaches parents to occasionally praise their children for not stuttering. Parents can be taught to say things like “That was lovely talking without bumps,” or “Good talking, no stuck words,” or something similar. It is essential for parents to do this in their own way. Every parent has a different style with a child, and different children need to be praised in different ways. It is also essential that parents are genuine with their praise and that they don’t do it excessively.

The second parent verbal contingency for stutter-free speech is *request self-evaluation*. This verbal contingency can be used when a child does not stutter for a certain period. That period can be as brief as a single utterance or as long as several hours. When no stuttering occurs for such a period, the parent can ask the child to self-evaluate stuttering during that period. The parent could say something along the lines of “Were there any bumps there?” and the expected response from the child would be “No.” Or a parent could say “Did you say all that smoothly?” and the expected response would be “Yes.”

The third verbal contingency for stutter-free speech is *acknowledge*. The difference between this and the previous two verbal contingencies is that the conversation is not paused at all. This is most important; the child’s everyday communication cannot be constantly disrupted each day by parent verbal contingencies. Also, acknowledging stutter free speech is different from praising stutter-free speech because it is a matter-of-fact statement rather than a positive comment. Examples would include: “That was smooth” and “No bumpy words.”

*Verbal contingencies for unambiguous stuttering*

These need to be introduced carefully because some children can be initially apprehensive about them. Also, verbal contingencies for stuttering are more likely to make children react negatively to the treatment than are contingencies for stutter-free speech. They are used less frequently than verbal contingencies for stutter-free speech. In other words, most of the verbal contingencies that children receive during the Lidcombe Program are for stutter-free speech. As is the case with verbal contingencies for stutter-free speech, every parent has a different style with a child, and different children will need to receive verbal contingencies for stuttering in different ways.

The first verbal contingency for unambiguous stuttering is *acknowledge*. As with the verbal contingency to acknowledge stutter-free speech, this verbal contingency needs to be not at all disruptive. The parent just notes that stuttering has occurred and moves on, saying something like “That was bumpy” or “That was a stuck word.”

The second verbal contingency for unambiguous stuttering is *request self-correction*. The parent asks the child to repeat the utterance without the stuttering moment. Mostly the child can do that, but if the child fails to do so, it is usually best for the parent to let it go. Examples of request self-correction would be to say, “Can you try that again” or “See if you can say that without the bump.” Request for self-correction occurs occasionally, not on the majority of or on most stuttering moments. The exception to this rule is when a child has only a few stuttering moments each day, which occurs towards the end of treatment.

*Optional parent verbal contingencies*

The Lidcombe Program Treatment Guide specifies two additional verbal contingencies that parents can use but which are optional. The first of these is *praise for spontaneous self-evaluation of stutter-free speech*. Older children with early stuttering receiving the Lidcombe Program will sometimes spontaneously self-evaluate their speech as stutter-free, saying something like “I didn’t do any bumps.” In such a case, a parent may respond with something like “Good boy, you’re listening for your smooth talking.”

The parent needs to be sure that the praise is for self-evaluation of stutter-free speech, not praise for stutter-free speech. Parents need to understand the difference between the two. For example, “Good boy, you’re listening for your smooth talking” is praise for self-evaluation of stutter-free speech, and “Good boy, that was smooth talking” is praise for stutter-free speech.

It is generally thought not to be a good idea to praise spontaneous self-evaluation of stuttered speech, such as “I just did a bump.” The reason for this is that it might confuse a child if the parent’s praise follows a moment of stuttering. If a child does spontaneously self-evaluate stuttering, parents can note that it occurred and tell the clinician at the next clinic consultation. Naturally, this is a desirable situation and a sign that the Lidcombe Program treatment process is working well.

The second optional verbal contingency is *praise for spontaneous self-correction*. When children correct a stuttered utterance without being asked by a parent to do so, parents can offer praise. Again, older pre-school children are the most likely to do this. The verbal contingencies that parents might use here include “Good girl, you fixed that bumpy word all by yourself,” and “You fixed that stuck word, good boy.”

**Some essential things about parent verbal contingencies***They are for unambiguous stuttering moments*

Lidcombe Program verbal contingencies for stuttering are for unambiguous stuttering moments. If parents have any doubt about a moment of stuttering, it is not a problem, and they can choose to not apply a verbal contingency. All children with clinical levels of stuttering will have many unambiguous stuttering moments each day, and parents will have plenty of them to work with. This normally only becomes a clinical issue at the end of Stage 1 when children have SR 0 or SR 1—*no stuttering* or *extremely mild stuttering*—during most days.

*Teach verbal contingencies for stutter-free speech first*

Clinicians don't teach parents how to do the verbal contingencies all at once. Normally, they first teach parents to do verbal contingencies for stutter-free speech so that children can become comfortable with the treatment. Then, they implement the parent verbal contingencies for stuttered speech with children when they are sure they are ready for it. It makes clinical sense to introduce verbal contingencies for stutter-free speech before verbal contingencies for stuttering, because it is an inherently positive approach.

*Be sure parents are doing them correctly*

The clinician needs to be sure that parents are doing verbal contingencies correctly, according to instructions. The way to do this is, at each clinic appointment, to have parents demonstrate exactly how they have been doing the verbal contingencies with the child during the previous week, and to give them feedback. This can be an imposing clinical task for junior clinicians. It involves watching parents give verbal contingencies, making constructive comments, and then demonstrating improvements with the child. However, it is essential to do this during clinic consultations. Otherwise, the treatment process will not work properly if parents continue to do verbal contingencies incorrectly.

*Verbal contingencies must be a positive child experience*

The Lidcombe Program treatment process will not work properly if verbal contingencies amount to a negative experience for the child. Verbal contingencies cannot be constant, intensive, or invasive. It is an essential clinical skill to identify when this is occurring during treatment, or even better, to identify when it might occur and prevent it. For some parents, it is necessary to introduce the treatment slowly and carefully so the clinician can be sure that the child is receiving supportive and enjoyable verbal contingencies. Otherwise, during clinic consultations, it will be obvious that the child is not happy with the treatment, and clinical outcome will of course be compromised.

**Verbal contingencies during practice sessions***Practice sessions*

The clinician teaches the parent to present verbal contingencies during practice sessions for 10–15 minutes, usually once per day, sometimes twice per day. Fewer or more each day can be recommended by the clinician as judged advisable. The parent typically sits with the child at a table, with suitable activities such as books and games. Such structure is not essential, however, and treatment during practice sessions can be done in many situations. But in many cases, perhaps most, the formality is useful.

*Their purpose*

Apart from parent training, the point of verbal contingencies during practice sessions is to accustom children to what the treatment procedures will be, and to focus their attention on the treatment target of “no stuttering.” Overall, verbal contingencies during practice sessions establish a positive experience of the Lidcombe Program for the child.

*Manipulating syntactic complexity and utterance duration*

Lecture One presented research evidence that stuttering increases with increasing syntactic complexity and utterance duration,<sup>†</sup> and that these findings have been replicated with children. Clinicians can use this information to teach parents to manipulate these variables when giving verbal contingencies

<sup>†</sup> Utterance duration is usually measured with words, syllables, or morphemes.

during practice sessions to minimise the occurrence of stuttered utterances, if needed. With such manipulation, treatment during practice sessions can involve giving the child a chance to respond to a range of utterance durations: from one- and two-word responses to several utterances. It will depend on the child's stuttering severity at the time of the activity. This parent manipulation of syntactic complexity and utterance duration is not a static procedure; parents change their utterance duration and language complexity as needed, according to the children's stuttering severity during the practice sessions.

## Verbal contingencies during natural conversations

### *Natural conversations*

When the clinician forms a view that it is appropriate, parents begin to judiciously introduce verbal contingencies during natural conversations. The natural conversations are everyday speaking situations with children: at mealtimes, during housework, in the bath, on the way to pre-school, in the park with the family, whilst shopping, and so on. Eventually, verbal contingencies during natural conversations will replace verbal contingencies during practice sessions, and the latter will not occur at all.



## Stage 2

### *The purpose of Stage 2*

There are three purposes of Stage 2. The first is to systematically hand over complete responsibility for management of children's stuttering to their parents. Second, Stage 2 is designed to detect any signs of impending relapse. As mentioned during Lecture Four, relapse after speech treatment for stuttering is common with adults. Although not so common with early childhood stuttering, it does occur after the Lidcombe Program. In fact, half the children in one report<sup>40</sup> showed some transient signs of stuttering a mean of 5 years after their treatment began. So, the third purpose of Stage 2 is a defence against relapse.

### *Treatment goals for Stage 2*

To progress to Stage 2, children need to meet the criteria mentioned earlier for two consecutive fortnightly appointments:

- (1) parent SRs of 0–1 during the week preceding the clinic appointment with at least four of those seven SRs being 0, (2) clinician SRs of 0–1 during the clinic appointment. (p. 10)<sup>24</sup>

### *Performance-contingent maintenance*

In 1980 the idea of a performance-contingent maintenance schedule was introduced to stuttering treatment, and its potential benefits were shown.<sup>41</sup> This amounts to the parent and child returning to the clinic and having to sustain treatment targets for increasingly longer intervals; two appointments 2 weeks apart, then two appointments 4 weeks apart, followed by the same thing at 8 and 16 weeks between appointments. If the child does not meet the Lidcombe Program treatment criteria at any appointment, the parent and child return to the start of the sequence. Stage 2 normally takes a year or more. The importance of following this procedure was shown in a report that half of children during Stage 2 fail to meet treatment criteria at least once during Stage 2.<sup>42</sup>

*A common Stage 2 problem*

When children attain the Lidcombe Program treatment criteria and there is no stuttering or nearly no stuttering, parents or clinicians, or both, can become complacent and not follow through with the prescribed Stage 2 maintenance program. This causes a risk that relapse will occur. The researchers who published a long-term clinical follow-up of the treatment<sup>40</sup> suggested that clinicians encourage parents to watch carefully for any signs of post-treatment stuttering during Stage 2. It is essential that verbal contingencies for stutter-free speech continue to occur during Stage 2, and that any unambiguous stuttering moments receive verbal contingencies from parents.

**Lidcombe Program problem solving**

Problem solving is a routine part of the Lidcombe Program. A study of common problems arose from 60 consultations with expert clinicians about cases where children were not improving.<sup>43</sup> Appendix Two at the end of this lecture shows the most common problems that needed to be solved. A more detailed and recent publication<sup>44</sup> presented 124 clinical challenges that occur during the Lidcombe Program treatment process and presented strategies to deal with each of those challenges.

**CLINICAL STRENGTHS AND LIMITATIONS OF THE LIDCOMBE PROGRAM****Strengths***Replicability*

The basis of the treatment process is replicable, with measurement and verbal contingencies clearly described in the Lidcombe Program Treatment Guide.<sup>24</sup> The guide specifies what occurs during each clinic appointment, and in what order. The benefit of a replicable treatment is that any properly trained clinician can be confident of doing it exactly the way it was demonstrated to be efficacious in clinical trials.

*Conceptual simplicity*

The essence of the treatment is that parents present five verbal contingencies to their children during practice sessions and natural conversations and measure their stuttering daily with a simple severity rating scale. Although the treatment is simple in concept, in practice it can be challenging to adapt it in a different way for every family, and to be sure that parents are doing the treatment correctly. These two features of the treatment—adapting it for each family and being sure that parents are doing it correctly—are essential for it to be successful.

**Limitations***It is not ideal for immediate early intervention*

The Lidcombe Program requires compliance from children. They need to participate in daily practice sessions and cooperate with the parent verbal contingency procedures. As noted in Lecture Two, many children begin stuttering prior to 30 months of age. If a clinician decides to begin treatment immediately with a child who has begun to stutter at that age, the Lidcombe Program may not be ideal. In fact, in the next lecture, treatment process research is discussed which shows that treatment times are longer for younger compared to older children who receive the Lidcombe Program.

*Safety issues*

As discussed in the next lecture, research has shown the Lidcombe Program to be safe. But it is still possible that occasionally a child could react negatively to verbal contingencies.<sup>45</sup> It is possible, therefore, that without proper clinician management to prevent such an event, a parent could give verbal contingencies in a negative and punitive manner that might be detrimental to a child's well-being. This does not occur often, but it can occur.

## TREATMENTS BASED ON MULTIFACTORIAL MODELS:

### I. PALIN PARENT-CHILD INTERACTION THERAPY<sup>†</sup>

#### Background

*A treatment based on a multifactorial model*

This treatment was developed at the Michael Palin Centre for Stammering Children in London. The treatment is one of many based on the multifactorial models described in Lecture Three. To reiterate briefly, these models state that what triggers stuttering and sustains it is predisposing motor, physiological, language, and developmental child variables and the way they interact with their living environments. None of these variables is *necessary or sufficient* for stuttering; they interact uniquely with the stuttering of each pre-school child. Palin Parent-Child Interaction Therapy is based on a theoretical position that is broadly consistent with this thinking.

The factors specifically mentioned by the developers of this treatment include:

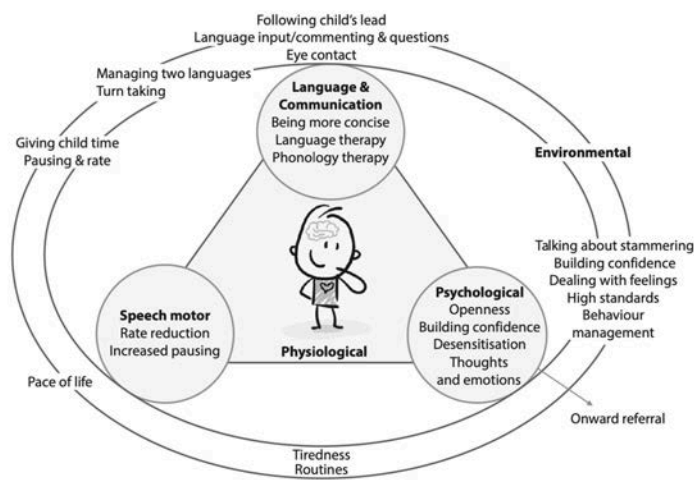
- (1) Psychological aspects such as child temperament and parent anxiety
- (2) Physiological factors such as gender, genetic history, and motor skill
- (3) Language development
- (4) Aspects of the living environment such as pace of life, communication and interaction style, parent language complexity, and rapid parent speech rate compared to that of the child.

Here again is the figure\* giving an overview the multifactorial model on which this treatment is based, which was presented during Lecture Three.

#### Other treatment influences

One of the developers of the treatment has noted that<sup>46</sup>

There is certainly an emphasis in identifying the individual child's strengths and needs, based on a belief that stuttering is multifactorial, heterogeneous and that the inherent vulnerability to stuttering is influenced by internal and external factors. The therapy itself is influenced by many approaches, including family systems theories, cognitive behaviour therapy, behaviour therapy, and solution focused brief therapy. (p. 3)



#### Clinical resource materials

Two journal reports contain overviews of the treatment.<sup>47,48</sup> A slightly longer overview appears in a book chapter,<sup>49</sup> and there is a comprehensive manual available.<sup>50</sup> Additionally, the Michael Palin Centre conducts a 3-day training for the procedure.<sup>51</sup> A web page at the Michael Palin Centre website contains general advice, in text and video media, to parents of children with early stuttering.<sup>52</sup> The advice to parents at that location includes the following:

<sup>†</sup> Thanks to Alison Nicholas at the Michael Palin Centre, London, for assistance with preparing this description of the treatment.

\* Adapted and reproduced with permission: Kelman, E., & Nicholas, A. (2020). *Palin parent-child interaction therapy for early childhood stammering*. Routledge. © 2024 Routledge.

“Having a short (5 minutes) one-to-one time with your child on a regular basis, when you are both calm and not in a rush and you are not likely to be interrupted

Thinking about your child's general well-being, his sleeping and eating habits, his health and his pace of life

Looking at your family's conversations - are you letting each other finish what you want to say? Is anybody hogging all the talking time? Do you interrupt each other when trying to speak?

Building your child's confidence by focusing on what he is doing well and praising him for this

Thinking about your child's language and whether he is trying to use sophisticated words and sentences to express himself. What kind of language are people using when they talk to him?”

## Overview

### *Pre-treatment assessment*

Palin Parent-Child Interaction Therapy begins with a detailed assessment that takes account of general speech and language development in addition to stuttering. The assessment is also designed to establish the extent to which children are aware of stuttering and how it may be affecting them socially and emotionally. This is needed because a key feature of the multifactorial model on which this treatment is based is that the putative factors responsible for stuttering combine uniquely for each child. In other words, the triggers for stuttering and what sustains it are different for every case of stuttering.

### *Diverse treatment goals*

A fundamental difference between this treatment and the Lidcombe Program is that Palin Parent-Child Interaction Therapy is not designed to achieve “no stuttering” or “nearly no stuttering.” Further, it has diverse goals. As stated by one of the developers of this treatment, “our aim is not zero stuttering during intervention. We seek to establish a decreasing trend in stuttering, reduced parental anxiety, and increased parental confidence in managing the stuttering” (p. 4).<sup>46</sup>

And, stated at another source:

The main focus of Palin PCI is the child, his or her profile of skills, and facilitating further development of the natural occurring fluency within the environment. It also aims to build on parents’ or caregivers’ knowledge and confidence in what helps and enhances existing behaviours that support fluency. (p. 69)<sup>49</sup>

And at another source:<sup>48</sup>

Palin PCI is explicit about the need to help parents address issues such as managing anxiety about stuttering, helping children manage emotions, confidence building, and other behaviour management such as setting boundaries and routines with, for example, sleeping, eating and turn taking. (p. 63)

### *Individual treatment design for families*

With Palin Parent-Child Interaction Therapy, the clinician forms a judgement about which of the factors, mentioned earlier, will be targeted in a treatment program. The clinician chooses from “interaction strategies,” “family strategies,” and “child strategies.”

<i>Interaction Strategies</i>	<i>Family Strategies</i>	<i>Child Strategies</i>
<i>Following child's lead</i>	<i>Managing two languages</i>	<i>Openness</i>
<i>Giving child time</i>	<i>Talking about</i>	<i>Building confidence</i>
<i>Pausing &amp; rate</i>	<i>stammering</i>	<i>Desensitisation</i>
<i>Eye contact</i>	<i>Building confidence</i>	<i>Thoughts &amp; emotions</i>
<i>Language input</i>	<i>Turn taking</i>	<i>Rate reduction</i>
<i>Commenting &amp; questions</i>	<i>Dealing with feelings</i>	<i>Increased pausing</i>
<i>Other (p. 109)</i>	<i>Tiredness</i>	<i>Being more concise</i>
	<i>High standards</i>	<i>Language therapy</i>
	<i>Behaviour management</i>	<i>Phonology therapy (p. 195)</i>
	<i>Routines</i>	
	<i>Pace of life</i>	
	<i>Other issues (p. 151)</i>	

The treatment manual<sup>50</sup> states

... most children require only the Interaction and Family Strategies component of the programme ... However, some children who continue to stammer may require further support with their response to their stammering, their confidence or their emotional well-being, as well as with direct speech modification strategies or with other skills to support their communication. Some children express that they would like more help with their talking and/or start to use strategies spontaneously. The Child Strategies teach the child what he can do to help himself. The Interaction and Family Strategies have created an environment in which the child is more likely to be able to use these Child Strategies. (p. 196)

### The treatment process

The treatment involves six weekly, 1-hour clinic visits, followed by a 6-week "consolidation" period, during which parents continue to implement strategies at home. Palin Parent-Child Interaction Therapy incorporates "special time," which is a 5-minute period that each parent spends individually with the child up to five times per week practising the "interaction strategies" above. These are video recorded in the clinic and discussed by the parents and clinician.

Although the treatment prescribes that there are six initial weekly clinic visits, there is some flexibility in allowing more if judged necessary. The developers state<sup>47</sup> that the duration of six clinic visits was selected because, at the time the treatment was originally developed, that number of clinic visits was a standard British National Health Service allowance to clinicians for treatment of children. The treatment manual states:

In the first instance, we will book six sessions and then you will practise at home for six weeks. You will then come back in for a review session, when we can decide if he needs any more therapy. For many children, all we need to do at that stage is see them from time to time to keep an eye on things. (p. 102)<sup>50</sup>

## TREATMENTS BASED ON MULTIFACTORIAL MODELS:

### II. RESTART-DCM TREATMENT

#### Background

RESTART-DCM<sup>†</sup> treatment has much in common with Palin Parent-Child Interaction Therapy, conceptually and procedurally. RESTART-DCM also has popularity in common with that treatment, being widespread in the Netherlands since the 1980s. In common with Palin Parent-Child Interaction Therapy, the treatment focuses primarily on strategies derived directly from multifactorial models of stuttering causality. However, as outlined below, it incorporates speech motor drills, speech

<sup>†</sup> This is an acronym for Rotterdam Evaluation Study of Stuttering Therapy-Demands and Capacities Model



restructuring techniques (see Lecture One), and verbal contingencies. Its current version<sup>53</sup> is derived from a method described in 1990.<sup>54</sup> Its developers recommend formal training in the method from the Netherlands.<sup>53</sup>

## Overview

Parents are informed as follows about the outcome of treatment:<sup>53</sup>

The best outcome of the therapy is when the child finds, feels and holds on to his or her “fluent speech groove”. However, there will always be a small number of children, whose Capacities are not adequate for achieving complete fluency. With the parents’ support, children can be prevented from developing negative feelings about their speech or about themselves, or from fighting against their stuttering. (p. 38)

After a comprehensive assessment, the treatment involves three phases. The purpose of Phase I is to reduce the demands on the child imposed by those in the child’s environment, and to reduce the child’s self-imposed demands. Phase II involves “training of the child’s Capacities for fluent speech” (p. 8) and is introduced if needed. Phase III is introduced if needed and involves training the child with direct methods for stuttering control, which are based on speech restructuring. During each of these three phases the following domains are dealt with: motoric, linguistic, emotional, and cognitive.

## Assessment

### *Parent assessment*

An initial parent interview includes (1) a medical and speech case history, (2) parent views about what caused the stuttering, (3) parent and child emotional reaction to the stuttering, (4) child temperament, (5) parent perceived stuttering severity and its burden, and (6) additional assessments thought to be necessary. The Demands and Capacities Model is explained briefly to parents.

### *Video interaction analysis*

A 15-minute video is made of a parent-child interaction, with both the parents and siblings included at the discretion of the speech-language pathologist. The video involves free play and puzzle activities. The video is then analysed using a Parent-Child Interaction Form, comprising motoric, linguistic, emotional, and cognitive features. The form contains 23 behaviours in the following categories. The treatment manual recommends transcription of two 5-minute samples of the video to assist categorisation:

- (1) Questions parent to child
- (2) Turn-taking behaviour
- (3) Parent response to stuttering
- (4) Parent(s) linguistic behaviour
- (5) Articulation rate
- (6) Other parental behaviour (p. 33)<sup>53</sup>

### *Child assessment*

All children are given “formalised standardized speech language testing, even if there are no concerns about his language development” (p. 10)<sup>53</sup> The speech and language tests are not specified. Additionally, children are given a measure of child attitude to communication (KiddyCat)<sup>55</sup> (see Lecture Twelve), an oral-motor assessment,<sup>56</sup> and the SSI-4<sup>57</sup> (see Lecture Four).

### *The first parent conference*

After assessment there is a parent conference without the child. The purpose of this is to (1) explain the Demands and Capacities Model in detail, (2) present assessment results, (3) introduce the “fifteen-minute parent-child Special Time,” and (4) introduce a treatment logbook.

## RESTART-DCM treatment

The treatment begins with 1-hour weekly consultations with the speech-language pathologist, parents, and child. The treatment manual states that:

The frequency of the sessions may vary throughout the treatment, depending on how it goes ... As a rule, after four sessions with parent and child, another parent interview is held at which the child is not present (p. 14)53

The structure of each clinical consultation is specified as follows: (1) the parent shows the clinician how modified behaviours were done during the previous week, (2) parents and clinician discuss the log book and progress during the previous week, (3) the clinician models a modified behaviour with the child, (4) the parent attempts the modification, (5) the clinician discusses the parent attempt, (6) the clinician discusses what is expected of the parent during the coming week.

### *Phase I: Reducing demands*

Based on the results of the assessment, one of many behavioural changes may be implemented under the categories of motoric, linguistic, emotional, and cognitive. Around 60 possible changes are outlined in the treatment manual, to be used as the clinician judges to be necessary. Some examples are:

#### (1) Reducing motoric demands:

Speaking with calm, relaxed and fluent speech movements in a natural manner with the child ... this will rarely exceed 3.5 syllables per second

Implementing long response time latency (approximately 1–2 seconds), definitely no overlapping in speech turn-taking

Maintaining normal eye contact, appropriate to the customs of the culture and the family.

#### (2) Reducing linguistic demands:

Ensuring a good balance between the number of spoken utterances per turn by the parent and by the child.

Primarily asking forced-choice or closed questions instead of questions in an open form

Modelling not always being able to think of the right name for something, and then describing it.

#### (3) Reducing emotional demands:

Understanding and acknowledging the child's temperament and adjusting his/her reaction to the child accordingly.

Modelling how to handle making a mistake in a relaxed way

Now and again, the parent will casually speak using the disfluencies/stutterings the child makes, with the aim of lessening the child's sensitivity to this.

#### (4) Reducing cognitive demands:

Posing a question in such a way that the child is free to comment or not

Trying to avoid 'Demand speech' as much as possible and to replace by a statement.

Following the child's initiative with his or her full attention instead of suggesting new subjects and asking the child to pay attention to these (pp. 16–20).

### *Phase II: Increasing the child's capacities*

The manual states that Phase II is introduced "if an insufficient level of fluency has been attained after completion of Phase I" (p. 21). That judgment is made by the speech-language pathologist on a case-by-case basis; there are no measurement-based criteria for the judgment.

If there is a need to increase the child's motoric capacities, based on the oral-motor assessment,<sup>56</sup> children receive an additional treatment: Speech Motor Training.<sup>58</sup> This treatment involves speech drills.<sup>†</sup> There is no indication in the RESTART-DCM manual about the treatment time needed for these drills. However, for older children, for whom it was developed, Speech Motor Training requires a mean of 8.1 clinical hours (standard deviation 8.8 hours).<sup>59</sup> In addition to clinical training, the parents practice the procedures for two 5-minute periods each day. Speech Motor Training is a 14-step programmed instruction drilling sequence involving syllable repetition. At each of the 14 steps, the clinician models the target, and the child repeats it. The programmed instruction procedure incorporates systematic increases in the number of syllables, number of syllables per breath group, and speech rate. The treatment incorporates praise and tokens. There is no indication in the research reports of RESTART-DCM treatment (see Lecture Seven) of what proportion of children receive speech motor training.

In addition to increasing the child's motoric capacities, some examples of other treatment agents introduced during Phase II are:

(2) Increasing the child's linguistic capacities:

Eliminating an eventual imbalance in the child's language profile (for example, relatively weak word comprehension) through appropriate language stimulation

Improving or automating sentence production through the automation of relevant sentence structures through play

Improving word retrieval by means of training word retrieval skills (p. 23).

(3) Increasing the child's emotional capacities:

Desensitisation to the experience of stuttering\*

... teaching the child skills to regulate his emotions himself

... the speech therapist creates moments during a treatment session in which a specific emotion is evoked in the child (p. 24).

(3) Increasing the child's cognitive capacities:

The concept of turn-taking/speech rules is taught

The child is taught concepts to enable him to 'talk about talking'

The child learns that 'mistakes are allowed' (p. 25).

*Phase III: Working more directly on fluency*

Phase III is implemented if "both the speech therapist and the parents feel that a further reduction in stuttering severity is possible" (p. 26). The procedures appear to be a variant of speech restructuring (see Lecture One): "a little bit slower, slightly less tense, with slightly more pronounced articulatory movement" (p. 26). These procedures are practiced during clinic consultations and at home with parents.

*Tapering off the therapy*

During this maintenance phase, clinical consultations are reduced to half an hour. The phase begins when the clinician and parents decide that "sufficient fluency has been attained" (p. 27). That decision may be made at the conclusion of Phase I, Phase II, or Phase III. The frequency of clinical consultations is gradually reduced. Parents gradually withdraw treatment procedures. The phase emphasises the need for parents to assume treatment responsibility and to resume treatment if relapse

<sup>†</sup> There are no published clinical trials of the Speech Motor Training procedure.

\* Procedures include verbal contingencies. As stated on page 24 of the manual: "a comment can be made in a calm voice about a stutter produced by the child ('Gosh, now your mouth got a bit stuck, same as me just now. Oh well, talking can be hard sometimes')."

occurs.

## CLINICAL STRENGTHS AND LIMITATIONS OF TREATMENTS BASED ON MULTIFACTORIAL MODELS

### Strengths

#### *They may not require child engagement*

Conceptually, with multifactorial model treatments the child does not have to do anything at all. It is only the parents who do the therapy, and the child is not required to engage with treatment. However, in practice, that is not what happens. For example, the RESTART-DCM treatment manual<sup>53</sup> specifies three Phases, only the first of which—“reducing demands”—requires no engagement from the child as parents change their behaviour. But the second phase of “increasing the child’s capacities” involves speech motor drills, and the third phase of “working more directly on fluency” seems to be a variant of speech restructuring adapted for pre-schoolers. Movement through the three phases is based on clinician judgment of need for such progression, and the one randomised trial of the treatment<sup>60</sup> (to be discussed in Lecture Seven) does not indicate which of these treatment components the children required. It is possible, though, that for some children the treatment might provide its intended effects with the first phase only.

### Limitations

#### *The importance of parent language*

Clearly, parent language makes a fundamental contribution to childhood development. Many clinical recommendations of multifactorial treatment models involve changes to customary parent language with children. Consideration of the merits of such strategies presents clinicians with a clinical judgment about their possible costs and possible benefits. During Lecture Seven, evidence in support of some parent language change for controlling stuttering is considered (reduced speech rate and increased inter-turn speaker latency), along with evidence against another (reduced questioning). As a background to their research about the latter issue, the researchers provided details about the importance of parent questioning for language development.<sup>61</sup>

#### *Potentially complex treatments*

Palin Parent-Child Interaction Therapy and RESTART-DCM treatment appear to be the most complicated and logistically challenging treatment for clinicians from among the three discussed during this lecture. The treatment manuals show around 60 therapy strategies are involved with each of the treatments. In addition to treatment methods derived from the Demands and Capacities model, RESTART-DCM incorporates speech motor drills, speech restructuring techniques, and verbal contingencies. With Palin Parent-Child Interaction Therapy, the “child strategies” of “rate reduction” and “increased pausing” are variants of speech restructuring. Data about the matter are limited, but one of the clinical trials of Palin Parent-Child Interaction therapy<sup>47</sup> suggested that, in practice, the treatment might be simpler than it appears at face value. In that trial, from four to six therapy strategies were chosen for each of the six families in the trial.

#### *Issues with the underpinning theoretical model*

A treatment based on a theoretical model of the nature of stuttering might be questioned if the model itself is questionable. As outlined during Lecture Three, there are grounds to argue that multifactorial models of early stuttering are indeed questionable, and consequently they have received considerable criticism. It seems fair also to state that these treatments are not straightforward applications of multifactorial models because Palin Parent-Child Interaction Therapy and RESTART-DCM treatment involve the clinical option of other techniques as needed, as discussed above: a variant of speech restructuring. Also, the latter treatment involves a speech motor training program if a child fails an oral motor assessment.

## THE WESTMEAD PROGRAM

### Background

#### *An old technique*

This treatment is currently in early developmental stages at the Australian Stuttering Research Centre, Sydney, Australia. It uses the well-known rhythm effect, or what is often called syllable-timed speech. As described during Lecture One, this is a fluency inducing condition that seems to have been used to treat stuttering centuries ago. It appears that the earliest documented modern use of this as a stuttering treatment occurred during the 1930s.<sup>62,63</sup> To summarise, when adults who stutter speak while they are saying each syllable to a rhythmic beat, either aided by a metronome or not, they stop stuttering. That is, until they stop speaking rhythmically, at which time stuttering resumes.

#### *Early application to early stuttering*

During the early 1980s some researchers looked for clinically useful effects when children spoke in rhythm.<sup>64</sup> The children in this report began speaking during each session with syllable-timed speech at 80–120 beats per minute, saying two-syllable words until they reached a target speech rate, which was from 104–112 beats per minute. Then, during each session, the children spoke in an increasing sequence from three to six single-syllable phrases, then conversational speech. During the last three sessions the rhythmic speech was phased out. The treatment was done solely within the clinic, with three visits per week for 5 weeks. The researchers concluded that the treatment was worthy of further investigation, but no subsequent reports were published.

#### *An intriguing experiment*

An experiment<sup>65</sup> involving 9–11-year-old boys showed that instructions were not necessary for them to decrease stuttering in the presence of a metronome. The researcher played a metronome in the background with a group of 20 children. Half of them were instructed to talk to the beat of the metronome and the other half received no instruction. Predictably, the children who were instructed to talk rhythmically did not stutter. But surprisingly, the study showed that the children who received no instruction also showed a significant treatment effect. In other words, the children showed a treatment effect from rhythmic stimulation without being instructed to speak that way. That was certainly suggestive of clinical value for syllable-timed speech with children.

### The Westmead Program treatment process

#### *Clinical resource materials*

These are located at the website of the Australian Stuttering Research Centre.<sup>66</sup> The *Westmead Program Treatment Guide* is a clinical reference for the treatment. There is a clinical severity rating (SR) chart for parents and clinicians: *Child Stuttering Severity Chart eForm*. That form is reproduced in Appendix One of this lecture. The website also contains an audio *Syllable-Timed Speech Training Model*, and four video *Syllable-Timed Speech Demonstrations*. There is also a *Stuttering Treatment Activity Guide*.

#### *Overview*

The Westmead Program directs parents to encourage children to use syllable-timed speech during everyday conversations. The aim is to achieve a typical speech rate and speech that does not sound unnatural in any way. For four to six times each day, for 5–10 minute intervals, the parent and child practise syllable-timed speech, and parents occasionally praise their child for using this speech pattern. Parents also prompt their children to occasionally use syllable-timed speech between these practice sessions. There are no set rules for how often these daily therapy activities should happen; the clinician makes a judgement for each child and family.

#### *Parents have a clinic consultation each week*

As with all evidence-based early childhood stuttering treatments, parents and children have a clinic

consultation each week. During each weekly consultation, the clinician teaches parents how to do the treatment and ensures that it is being done properly.

#### *Treatment goals during Stage 1 and Stage 2*

As with the Lidcombe Program, Westmead Program treatment criteria are “no stuttering” or “nearly no stuttering” for a long time. The goal of Stage 1 is to achieve no stuttering or nearly no stuttering, and the goal of Stage 2 is for that to be sustained for a long time. As with the Lidcombe Program, Stage 2 of the treatment is sometimes referred to as maintenance. The treatment criterion measure is the SR scale, which is used also with the Lidcombe Program. Stage 1 concludes when these treatment criteria are met:

To progress to Stage 2, the following criteria need to be met for two consecutive fortnightly consults: (1) clinician SR of 0 or 1 during the consultation, and (2) daily parent typical SRs of 0-1 during the week preceding the consultation, with at least four of those seven SRs being 0. A minimum requirement during Stage 2 is for parents to document SRs during the week preceding the consultation. However, the clinician may request parents to document SRs more often. (p. 6)<sup>24</sup>

#### *Stage 1*

Stage 1 of the treatment has two components, Stage 1A and Stage 1B. During Stage 1A, the parent and child attend the clinic for 30–60 minute sessions so they can both learn to do the syllable-timed speech pattern. During this period the parent and child establish a routine where syllable-timed speech is practiced each day. The clinician teaches the parent, where necessary, to modify utterance duration and grammatical complexity to make syllable-timed speech easier to learn. Generally, children learn to do the speech pattern quickly and can do it during conversation during the first few sessions. Then the clinician directs the parent to have the child attempt it during conversations between practice sessions.

Stage 1B begins when the parent and child are practising and using syllable-timed speech during the day correctly. As with the Lidcombe Program, it is critical to be sure that parents are doing what the clinician intends. Fortnightly consultations begin during Stage 1B.

#### *Stage 2*

When children attain the treatment criteria, Stage 2 begins, and the family has clinic consultations less frequently during a period of 1 year. During Stage 2, parents are instructed to gradually stop doing the practice sessions each day. If, during a Stage 2 clinic consultation, the child does not meet treatment criteria, the clinician has the option of either stopping progress through Stage 2, while the problem is resolved, or to return the child to Stage 1 to re-establish treatment gains.

## **CLINICAL STRENGTHS AND LIMITATIONS OF THE WESTMEAD PROGRAM**

### **Strengths**

#### *A simple procedure*

Of all the treatments discussed in this lecture, the Westmead Program is the simplest. Speaking with syllable-timed speech seems to be easy for children to learn. So much so, in fact, that as soon as the parent and child learn to do the procedure, clinic consultations begin to occur fortnightly.

#### *It may be useable for immediate early intervention*

Rhythmic stimulation is quite a simple procedure, so it may be more useable with younger children than is the case for the Lidcombe Program.

#### *Treatment credibility and expectancy*

There is a strong theoretical basis to the Westmead Program, not in the sense of stuttering causality, but in terms of the mechanism that might explain how it should work. Apart from the fact that syllable-timed speech seems to be the oldest stuttering treatment method on record, the P&A Model described

during Lecture Three provides a credible explanation for how it might work; syllable-timed speech removes the stress contrasts that trigger stuttering moments.

### **Limitations**

#### *A repetitive and drill-like procedure*

This aspect of the treatment could prove to be troublesome as it develops with further clinical trials. Even though parents rapidly learn to do the treatment with their children, it may prove to be quite wearying for them to sustain for long periods to obtain durable stuttering control.

### **SUMMARY**

The early years of stuttering are a time when it is at its most tractable. At that time parents have optimal contact with their children during daily life. Therefore, early childhood stuttering intervention is a desirable clinical option. There are three treatment types for children with early stuttering for which there is clinical trial evidence: the Lidcombe Program, treatments based on Multifactorial Models, and the Westmead Program. The three treatments differ in clinical processes, and each has unique strengths and limitations.





**APPENDIX TWO**

Common Lidcombe Program problems based on consultations with expert clinicians about cases where children were not improving.<sup>43</sup> The percentage of consultations for which the problem occurred is presented in the third column.

<i>Speech measures</i>	<i>SRs not collected or collected infrequently. Parents unreliable using SRs.</i>	14%
<i>Training parents</i>	<i>Parents presenting verbal contingencies incorrectly. Family members presenting contingencies without training.</i>	14%
<i>Practice sessions and natural conversations</i>	<i>Treatment during practice sessions used for too long into treatment. Treatment during natural conversations introduced too early.</i>	13%
<i>Verbal contingencies for stutter-free speech</i>	<i>Parents not presenting enough of them. Used during practice sessions but not during natural conversations.</i>	9%
<i>Verbal contingencies for unambiguous stuttering moments</i>	<i>Parents using them excessively. Parents presenting them in a manner that children don't like. Parents presenting them inaccurately.</i>	8%
<i>Low rate of verbal contingencies</i>	<i>Verbal contingencies given infrequently during practice sessions and conversations. No verbal contingencies given at all.</i>	8%
<i>Child has other speech or language problems</i>	<i>Clinician concurrently manages several different conditions.</i>	8%
<i>Stage 2</i>	<i>Entry to Stage 2 without attaining treatment criteria. Stopping verbal contingencies during Stage 2.</i>	5%
<i>Missing components of Stage 1</i>	<i>Weekly 45–60 minute clinic consultations. Parent training with verbal contingencies. Consistent application of treatment.</i>	5%
<i>Stuttering severity fluctuates</i>	<i>Clinicians unaware this is common. Problems measuring treatment progress. Clinicians not aware it could be caused by treatment or natural variability.</i>	5%
<i>Stuttering twins being treated</i>	<i>Clinician uncertainty about treating concurrently or separately. Expectations about treatment times.</i>	2%
<i>Sensitive child</i>	<i>Managing dislike of verbal contingencies.</i>	2%
<i>Parent scepticism</i>	<i>Managing parent confusion about treatment and doubt about its benefits.</i>	2%
<i>Child unaware of stuttering</i>	<i>Clinicians uncertain about whether to make child aware of stuttering before treatment.</i>	2%
<i>Problematic parent-child relationship</i>	<i>Parent focused negatively on stuttering rather than constructive treatment.</i>	1%

†

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## LECTURE SEVEN: THE EARLY CHILDHOOD STUTTERING INTERVENTION EVIDENCE BASE

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### CLINICAL TRIALS OF ONE TREATMENT

#### The Lidcombe Program

##### *In-clinic Phase I-III trials*

The Lidcombe Program was developed for the traditional format of weekly clinic visits. For this format, the first Phase I non-randomised clinical trial with Australian children was published in 1990.<sup>1</sup> Subsequently, there has been a Phase I nonrandomised trial with Malaysian children<sup>2</sup> and one with Kuwaiti children.<sup>3</sup> There have been three non-randomised Phase II trials with Australian children,<sup>4,5,6</sup> and one Phase III randomised controlled trial with New Zealand children.<sup>7</sup> One report<sup>8</sup> involved 3–7-year follow-ups of the children treated in that trial. One of the Phase II trials<sup>6</sup> began as a randomised controlled trial, but the researchers could not retain the control group, so it finished up as a Phase II trial with just the children who completed the Lidcombe Program.

The Phase III randomised controlled trial<sup>7</sup> recruited 54 New Zealand children, 12 of them girls, and randomised 29 of the children to a Lidcombe Program arm and 25 to a no-treatment control arm. Two children dropped out of each arm. The primary outcome was percentage syllables stuttered (%SS), measured in three everyday childhood speaking situations at pre-randomisation, and again at 3, 6, and 9 months post-randomisation. At 9 months post-randomisation, %SS for the Lidcombe Program arm was 1.4 and 3.9 for the control arm. This result was statistically and clinically significant.

A Phase III randomised controlled trial<sup>9</sup> compared a standard treatment arm to a group treatment arm involving three families per group. The latter treatment arm involved a “rolling group” model, where a new family entered the group each time a family left the group. Fifty-four children were randomised, and clinical outcomes for the standard and group treatment arms were consistent with outcomes from other clinical trials. However, the children in the group arm required around half the number of clinical hours than the children in the standard arm. Therefore, the group Lidcombe Program treatment model was clinically efficient, although the treating clinicians in the trial “found group treatment to be more taxing but clinically gratifying” (p. 1606).<sup>9</sup>



A Phase II randomised trial<sup>10</sup> with three arms compared the traditional weekly visit treatment format with two alternatives: two clinic visits each week, and one clinic visit each two weeks. Thirty-one children were randomised to one of the three service delivery models. The conclusions that can be drawn from this study are limited because of its preliminary nature, having low participant numbers and high dropout rates; six, seven and eight children remained in the three treatment arms at 9 months post-randomisation. However, considering this, results showed no evidence of any difference for %SS scores at that assessment. Generally, it seemed that two clinic visits each week was not a feasible model for practical reasons. Despite the varying number of clinic visits per week, the median number of weeks to complete Stage 1 was similar for the groups. Of particular interest was the similar number of weeks needed for the groups, with a clinic visit each week and one visit fortnightly: 23 and 24 weeks, respectively. The authors concluded that, considering the health economics of the matter, further clinical trialling is warranted. However, no further trials have been published.

*Telehealth Phase I-II trials*

Telehealth was considered in detail during the previous lecture: technology used to treat clients when they are not in the clinic. There have been three low-tech telehealth trials of the Lidcombe Program with Australian children using the telephone: two Phase I trials<sup>11,12</sup> and one randomised Phase II trial.<sup>13</sup> A Phase I trial of video telehealth has also been published.<sup>14</sup> A Phase III randomised controlled telehealth trial has been published as well, with an in-clinic and a telehealth arm.<sup>15</sup> The design is known as a parallel, open plan, non-inferiority randomized controlled trial. Results showed no reason to believe that the telehealth Lidcombe Program was less efficacious in terms of stuttering severity outcomes, or cost, than the clinic presentation. In fact, the telehealth arm of the trial had clinical consultations that were 17% shorter than the clinic arm. There was no reason to believe that parents and children in either arm of the trial had a different relationship with the treating clinicians. For both treatments, there was a general association between stuttering severity and parent satisfaction with fluency.<sup>16</sup> A clinical guide to conducting the Lidcombe Program by telehealth is available.<sup>17</sup>

It is not clear at present where telehealth Lidcombe Program developments will lead. It could turn out that this treatment method will be suitable for most families. On the other hand, this may not be so, and the final place for telehealth Lidcombe Program intervention may be as part of a stepped care public health approach to early stuttering, as described during the previous lecture.

*The Lidcombe Program in different cultures*

A systematic review<sup>18</sup> identified eight data-based studies of the treatment in non-English speaking countries. The languages involved were Arabic (Kuwait), Baluchi and Persian (Iran), Dutch (The Netherlands), Swedish (Sweden), China and Malaysia (Mandarin), and Bulgaria (Bulgarian). The review concluded that the treatment is efficacious in different cultures and languages, although it can take longer to complete than in English. The review concluded that it

... can meet the needs of bilingual children and families and seems to be deliverable in a multilingual context even when the service deliverer and the child do not speak the same language. (p. 12)

The treatment focusses on being a positive experience for children, and, as such, praise and acknowledgment for stutter-free speech is usually a clinically essential parent verbal contingency. However, when formulating a Phase I trial of the Lidcombe Program in Malaysia,<sup>2</sup> the researchers pointed out that the treatment was developed for Western cultures. Based on a study of Malaysian parents and children with early stuttering,<sup>19</sup> they concluded that “praise and acknowledgment of desirable behaviours ... appear to be used only infrequently in Malaysian cultures, and that when they occur, may not be varied in expressions” (p. 30).<sup>2</sup>

Four Malaysian children with early stuttering were studied, one of whom was treated in Mandarin Chinese and the others in English. Based on beyond-clinic recordings 12 months after Stage 1, one child had %SS scores of zero, and another child had scores below 1.5 %SS. The third child had scores around 3.0 %SS and the fourth child did not reach Stage 2. The numbers of clinic visits to reach Stage 2 were 21, 31, and 57, which were longer than usual treatment times for the Lidcombe Program (to be reviewed shortly). The researchers reported that this seemed to have been caused by additional time required to teach the parents verbal contingencies, particularly praise for stutter-free speech. The researchers suggested approaches to the cultural issues about praise with the Lidcombe Program, such as variation of tonal and facial expression.

Similar themes emerged during a Phase I trial with six Kuwaiti children with early stuttering.<sup>3</sup> Four of the children completed Stage 1 and, based on beyond-clinic recordings, were stuttering below 1.0 %SS during Stage 2. The authors reported that praising the children did not come naturally to the parents, and more time was spent training them to use verbal contingencies than is typical for Western parents. Additionally,

Cultural factors were evident in the current study, such as the inability for women from traditional Bedouin families to drive to sessions on their own and

relying on their husbands and other family members for transport. Other reasons for missing sessions included religious holidays ... (p. 230)

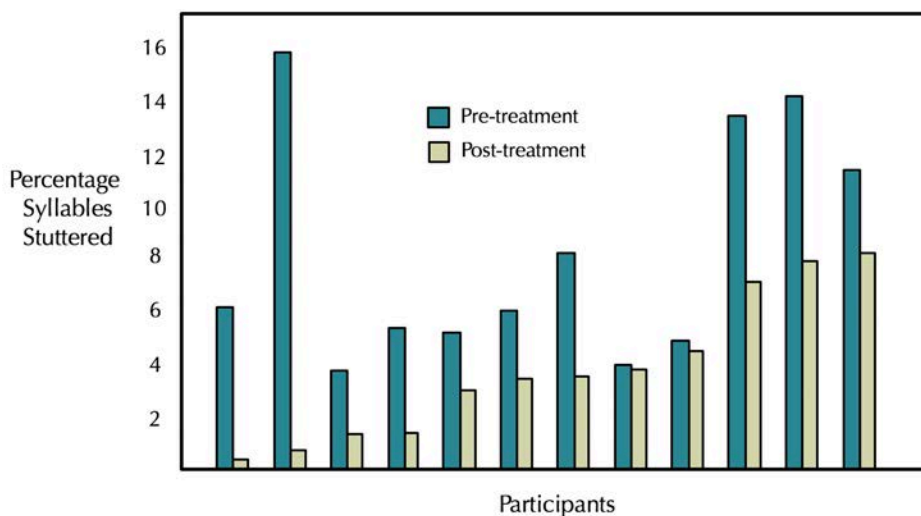
#### *The Lidcombe Program with co-occurring speech sound disorder*

A Phase I trial<sup>20</sup> with five boys, ages 3–4 years, involved treatment for stuttering with the Lidcombe Program concurrently with treatment for speech sound disorder. The children were assessed at pre-treatment, at entry to Stage 2 of the Lidcombe Program, and at 9 and 12 months after the start of treatment. The primary stuttering outcome measure was %SS based on two 10-minute conversation samples in everyday situations. Four of the children completed Stage 1 in 14–22 clinic visits, which is consistent with clinical benchmarks (to be discussed shortly). One child did not complete the treatment. Pre-treatment stuttering for the four children was in the range 2–15 %SS, and at 12 months post-treatment they were all below 1.0 %SS. At 12 months post-treatment, all children had shown clinically significant improvement with speech sound disorder to within developmental expectations. The authors concluded that “young children with co-occurring stuttering and speech sound disorder may be treated concurrently using direct treatment approaches” (p. 251).<sup>19</sup>

### Palin Parent-Child Interaction Therapy

#### *In-clinic Phase I trials*

The developers of this treatment have reported two Phase I clinical trials of it using in-clinic service delivery, with a total of 12 children.<sup>21,22</sup> The first trial<sup>21</sup> recruited nine families, of whom three dropped out, and the latter trial<sup>22</sup> recruited six children who were retained in the trial. The results across the two non-randomised trials are presented in the figure below for the 12 children. For some of the children in the figure, follow-up data are for 6 months post-treatment,<sup>22</sup> and for some the follow-up data are 12 months<sup>21</sup> post-treatment.



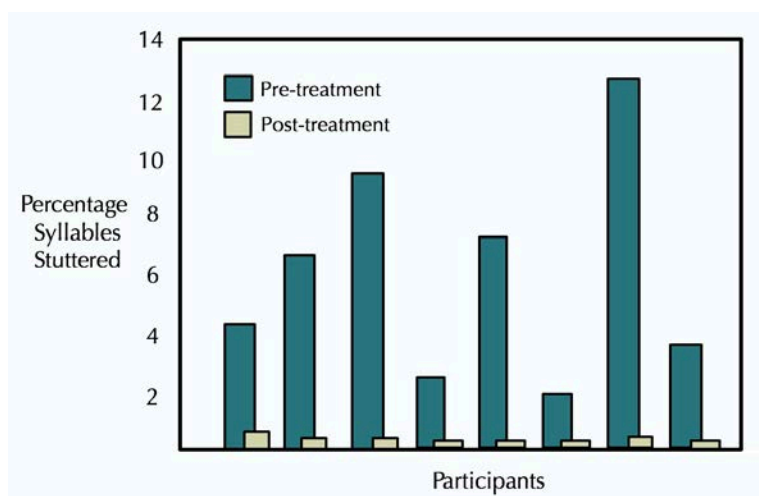
The first four children, on the left of the graph, achieved stuttering reductions to around 1.0 %SS or lower, but this was not the case for the other eight children. Two children showed almost no stuttering reduction. Overall, the pre-treatment to post-treatment reductions of %SS were 64% for the 12 children. Considering that non-randomised designs overestimate effect sizes, this result could arguably reflect natural recovery. These data need to be interpreted keeping in mind that no stuttering or nearly no stuttering is not a goal of Palin Parent-Child Interaction Therapy.



## The Westmead Program

### *In-clinic Phase I-II trials*

After Phase I trials,<sup>23,24</sup> a Phase II trial<sup>25</sup> recruited 17 children. Only eight of these children completed the treatment. With that caveat in mind, along with the caveat about interpreting non-randomised evidence, the corresponding figure here suggests that the treatment may have some merit. For the eight children who completed the trial, the mean post-treatment score at entry to Stage 2 was 0.2 %SS.



## CLINICAL TRIALS COMPARING TWO OR MORE TREATMENTS

### Lidcombe Program compared to RESTART-DCM treatment

#### *Method*

This is the largest randomised controlled trial reported for any stuttering treatment,<sup>26</sup> randomising 99 children to a Lidcombe Program arm and 100 children to a RESTART-DCM arm. The trial was in The Netherlands, with the treatments presented in Dutch. To be eligible for the trial, children were required to have been stuttering for at least 6 months and to be stuttering more severely than 3.0 %SS. The children randomised to the Lidcombe Program arm had a mean age of 51 months, and children randomised to the RESTART-DCM arm had a mean age of 52 months. Children in the former group were treated with a version of the Lidcombe Program treatment guide available at the time,<sup>27</sup> and children in the latter group were treated with the RESTART-DCM manual mentioned during the previous lecture.<sup>28</sup> Children were followed up for 18 months after the start of treatment. A detailed methodological evaluation of the RESTART trial has been published.<sup>29</sup>

#### *Treating clinicians*

Twenty-four clinicians at 20 clinics throughout the Netherlands treated the children. All clinicians received training from the Lidcombe Program Trainers Consortium<sup>30</sup> and “DCM based treatment training is included in the regular clinical education in the Netherlands” (p. 3).<sup>26</sup> Clinicians had a mean of 3.7 years of experience with the Lidcombe Program and a mean of 15 years of experience with the RESTART-DCM treatment. The researchers reported various strategies designed to maximise treatment fidelity, including 3-monthly clinician meetings and clinician treatment logs. The experiences of the treating clinicians are described in a separate publication.<sup>31</sup>

#### *Primary outcome*

The primary outcome was “the percentage of nonstuttering children at 18 months, operationalized as  $\leq 1.5\%$  syllables stuttered” (p. 4).<sup>26</sup> This measure was derived from %SS, at 18 months after the start of treatment, measured from three 10–15 minute audio recordings of the children during a period of 2 weeks. Two of these recordings were during conversations with parents and others at home, and one was with a non-family member outside the home. At 18 months after the start of treatment, 28% of the children in the Lidcombe Program arm had not completed Stage 2 of their treatment, and 35% of the children in the RESTART-DCM arm had not attained final treatment targets.<sup>32</sup> There were 21 dropouts (11%): nine in the RESTART-DCM arm and 12 in the Lidcombe Program arm. It is not stated in the report, but, for the 72 children who completed Stage 1 in the Lidcombe Program arm, the mean number of clinic sessions was 16.4 and the median number of treatment sessions was 14.5.<sup>33</sup>

### Secondary outcomes

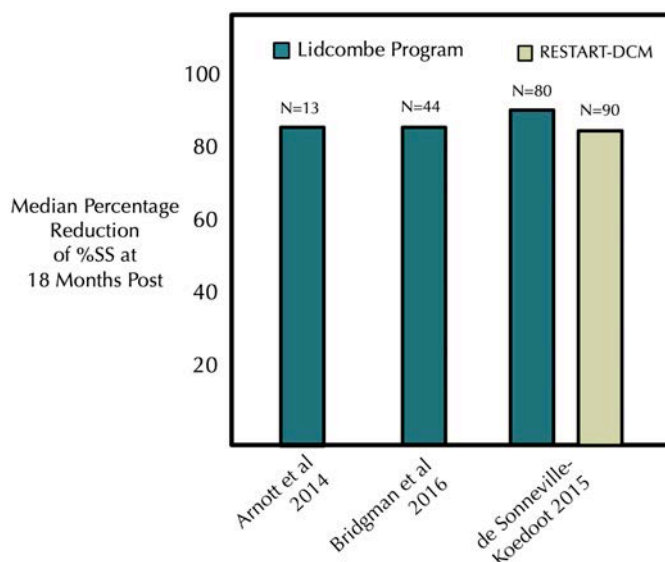
A range of secondary outcomes was reported, including %SS and parent and clinician severity rating with an 8-point scale. These were reported for pre-treatment, and 3, 6, 12, and 18 months after the start of treatment. It is not made fully clear in the report, but the %SS measures at pre-treatment, and 3, 6, and 12 months after the start of treatment, were based on the same methods as the %SS measures at 18 months after the start of treatment: three audio recordings of the children beyond the clinic.<sup>34</sup> Additional secondary outcomes were a health-related quality of life measure (EQ-5D),<sup>35</sup> a measure of child attitude to communication (KiddyCat)<sup>36</sup> (see Lecture Twelve), and three measures of child emotional and behavioural problems derived from the Child Behavior Checklist.<sup>37</sup>

### Primary outcome results

At 18 months after the start of treatment, 86 children remained in the Lidcombe Program arm, and 91 remained in the RESTART-DCM arm. There were 76.5% of “non-stuttering” children in the Lidcombe Program group and 71.4% of “non-stuttering” children in the RESTART-DCM group. These differences were not significant. Results remained nonsignificant when the cut-off %SS scores for “non-stuttering” were changed to 1.0 %SS and 2.0 %SS.

In terms of the 95% confidence intervals (see Lecture Five), that were reported, it is appropriate to interpret these results as showing no evidence of a difference between the treatments, rather than evidence that the treatments are equivalent. For %SS scores 18 months after the start of treatment, the mean difference between treatments was 0.3 %SS, with a 95% confidence interval for the difference of -0.4–0.9 %SS. That confidence interval contains zero (see Lecture Five). Additionally, for the percentage of “non-stuttering” children at 18 months after the start of treatment, the 95% confidence intervals were 66–84% for the Lidcombe Program and 61–80% for RESTART-DCM. Arguably, the range of these confidence intervals includes differences that are clinically significant.

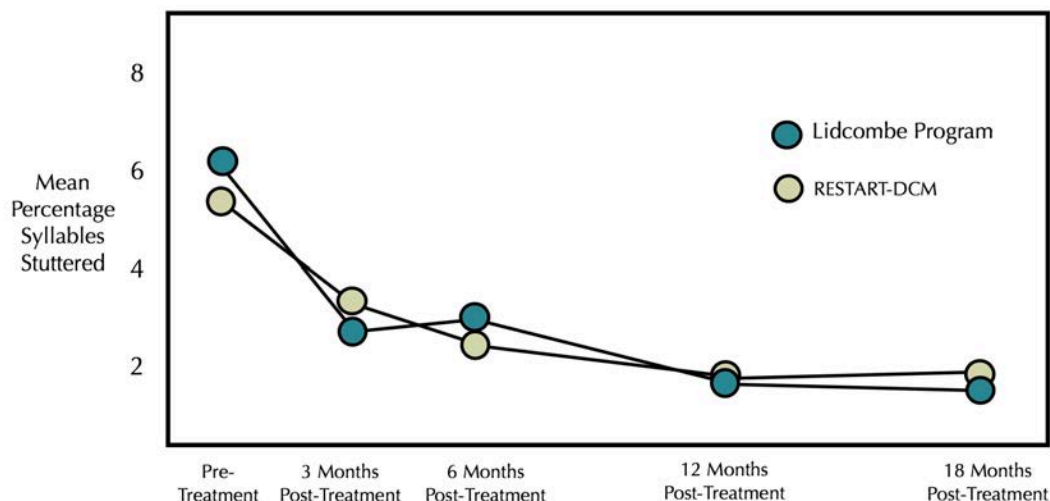
It would be useful to have some way of comparing the outcomes of the RESTART trial with the outcomes of the dedicated Lidcombe Program clinical trials discussed earlier. Any such comparison needs to be guarded because the trials concerned were conducted in different countries, at different times, and with different research protocols. There is also the problem of possible—even likely—differences of %SS scores by clinicians in different countries, as discussed during Lecture Four. With that in mind, the figure presents median percentage reductions for the RESTART trial and the data from the two standard treatment arms from Lidcombe Program randomised trials at 18 months post-randomisation.<sup>9,15</sup> With the caveat that such a comparison needs to be guarded, the data in the figure suggest no evidence that outcomes in terms of stuttering reductions are different across the three trials.



### Secondary outcome results

The paper reported that “most outcome measures were slightly in favour of the direct approach (LP), but the few significant interaction terms were deemed negligible due to their small effect sizes” (p. 11).<sup>26</sup> For the entire 18-month pre-treatment to post-treatment period there was a statistically significant effect favouring the Lidcombe Program for %SS and parent severity rating (see Table 2, p. 8–10), but with small effect sizes. In other words, reported effects were statistically but not clinically

significant (see Lecture Five). There were no significant changes pre-to post-treatment for the quality of life measures. With the Lidcombe Program group, there were significant post-treatment improvements for the three measures of emotional and behavioural problems, but these were attributable to a pre-treatment difference between the groups. For the attitude to communication scores, there was an almost statistically significant (unadjusted  $p=.06$ ) post-treatment improvement for both groups. The figure\* presents the %SS measures for the trial at pre-treatment, and 3, 6, 12, and 18 months after the start of treatment.



#### *An economic evaluation of the two treatments*

An economic evaluation of the RESTART clinical trial<sup>38</sup> reported that, at 18 months after the start of treatment, health outcomes were slightly better for the Lidcombe Program than for the RESTART-DCM treatment. One measure attained statistical significance with a small effect size (Cohen's  $d=0.17$ ): quality adjusted life years. The authors concluded that "cost-effectiveness and cost-utility ratios were in favour of the LP. The LP is considered a good alternative to RESTART-DCM treatment in Dutch primary care" (p. 106).<sup>38</sup>

#### *A critique*

The RESTART trial attracted a negative critique<sup>39</sup> pointing out that it was without a control group that received no treatment, hence that it presented "no value for clinical management because the treatments investigated were not shown to be more effective than no treatment" (p. 65). The treatment was also criticised because of the paradoxical criterion of  $\leq 1.5$  %SS as non-stuttering.<sup>†</sup> The authors responded<sup>40</sup> by conceding that the RESTART trial could not determine whether either treatment was better than natural recovery, but pointed out that the goal of the study was not to do that. In relation to the issue of the criterion for non-stuttering, the authors reiterated that the results of the trial were identical when different criteria of  $\leq 1.0$  and  $\leq 2.0$  %SS for non-stuttering were applied.

\* Adapted and reproduced with permission: De Sonnevle-Koedoot, C., et al. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PLoS One*, 10, e0133758. © 2015 de Sonnevle-Koedoot et al..

† That criticism about the primary outcome seems reasonable. For example, at a childhood speech rate of 200 syllables per minute,  $\leq 1.5$  %SS represents up to 180 stuttering events for every hour of speech. Clearly, that cannot be described as "non-stuttering." It is arguable that a simpler and more interpretable primary outcome for the trial would have been the gold standard of post-treatment %SS scores compared across treatments groups, as is routinely used in clinical trials.

## Lidcombe Program compared with two Westmead Program versions

### *Background*

A clinical trial<sup>41</sup> was conducted prompted, by the limitations of the Lidcombe Program that were discussed during Lecture Six, and prompted by the potential advantages to the Westmead Program as outlined in that lecture. Additionally, the authors argued that the Westmead Program is potentially useful because it does not require the dedicated practice sessions required with the Lidcombe Program, and that non-randomised clinical trial data suggest that it may require fewer treatment hours. Two versions of the Westmead Program were devised for the trial, one incorporating the verbal contingencies of the Lidcombe Program. The rationale for this was a high dropout rate in a previous trial and a suggestion “that families tended to withdraw from treatment at the point when low-level stuttering severity had been attained but not stabilized” (p. 507).<sup>41</sup> The report raised the idea that “for such cases, the final stages of clinical progress need to be hastened with the addition of contingencies for stuttered and stutter-free speech” (p. 507).

### *Method*

The trial was a three-armed randomised controlled trial with the Lidcombe Program as the control group and the two Westmead Program versions as the experimental groups, one with and one without verbal contingencies. There were blinded outcome assessments at 9 months post-randomisation. There were 91 children recruited, 33 to the Lidcombe Program arm and 28 and 30 to the two Westmead Program arms. This was the first clinical trial of either treatment without a lower age restriction; children of any age who were stuttering were eligible to be participants.

### *Treating clinicians*

The treatments were conducted in Melbourne and Sydney, Australia, in two community and two university research clinics. All clinicians had received Lidcombe Program training from the Lidcombe Program Trainers Consortium and received Westmead Program training from its developers.

### *Primary and secondary outcomes*

The primary outcome was %SS measured at 9 months post-randomisation from two 10-minute recordings of the children. One of those involved the children speaking to a non-family member outside the home, and the other with an adult family member at home. The secondary outcome was the number of clinic visits required to complete Stage 1.

### *Results*

No evidence was found of any difference in %SS scores between the groups at 9 months post-randomisation. There was evidence to support earlier trials that treatment times were shorter for the Westmead Program, with a median of 30 clinic visits to complete Stage 1 of the Lidcombe Program and only 18 and 16 visits for the Westmead groups.

### *Limitations*

A major limitation of this study is that, although the authors attempted to fix the Westmead drop-out problem with an adapted Westmead Program, they failed to do so. The drop-out rates for both Westmead arms were 43%, and there was also a substantial Lidcombe Program drop-out rate of 27%. This weakens the confidence that can be placed in the results, even though a statistical technique called multiple imputation was used to adjust for the problem.

The authors argued that some novel aspects of the trial may have accounted for dropouts. There were no speech and language exclusion criteria, and 13 of the children were younger than 3 years, which has never occurred previously in a clinical trial of early stuttering intervention. It was also the first trial of either treatment involving community clinics. Regardless, the authors concluded that “parents and children may simply find the treatment boring” (p. 13).<sup>41</sup> The authors also stated that they are attempting to deal with this problem by developing a standalone internet version of the Westmead Program.

## CLINICAL TRANSLATION

### The Lidcombe Program

One translational study,<sup>†</sup> an in-clinic Phase IV trial,<sup>42</sup> has explored whether the results of Lidcombe Program clinical trials can be achieved in clinical communities. The study involved 31 Australian community clinicians who treated 57 children with early stuttering. Outcome measures were %SS during everyday childhood conversations at 9 months after the start of treatment. Statistical regression modelling was used to determine whether any variables could predict that outcome: (1) pre-treatment stuttering severity, (2) speech or language disorders in addition to stuttering, (3) whether the clinicians had received training from the Lidcombe Program Trainers Consortium,<sup>30</sup> (4) the duration of weekly clinic visits, and (5) the mean period between clinic visits. At 9 months post-treatment, 12 children (21%) had withdrawn from their treatment, 47 (65%) had completed Stage 1, and eight (14%) were still in Stage 1.

The mean 9 months post-treatment stuttering severity for all the children was 1.7 %SS. However, Consortium training was a significant predictor of outcome. Children treated by Consortium trained clinicians attained a mean of 1.1 %SS at 9 months post-treatment and those treated by clinicians without such training scored a mean of 2.4 %SS, which is more than double. No other predictors of outcome were found. The authors concluded that, for clinicians with Consortium training, Lidcombe Program community outcomes can match those attained in clinical trials.

That translation study was followed up with a replication and extension<sup>43</sup> involving more participants and including English community clinicians: 36 clinicians in Australia and 15 in England, and 121 of their stuttering pre-school clients and families. Again, the community clinicians matched the outcomes reported in Lidcombe Program clinical trials. However, the English clinicians attained outcomes around 1.0 %SS higher than the Australian clinicians, although that effect was not reflected in parent severity rating scores. Therapist drift emerged as an issue in this report, and the authors suggested that it may be related to the pressures of community health care. The previous finding about Consortium training as an outcome predictor<sup>†</sup> were not replicated, leading the authors to conclude that standard professional preparation or specific Lidcombe Program post-graduate training may be sufficient for treatment translation. The result may be generalisable to other countries besides England and Australia.

A study involving six community clinicians<sup>44</sup> evaluated the cost-effective rolling-group Lidcombe Program model used in a previous randomised trial.<sup>9</sup> Participants were 19 children with early stuttering, mean age 49.1 months, treated by six generalist clinicians in four Australian rural towns. Within-clinic measures of %SS were collected at pre-treatment and at 6 and 9 months after the start of treatment. Percentage syllables stuttered scores were 7.4 at pre-treatment and 1.4 and 1.3 at the 6 and 9 months post assessments, respectively. Those results were obtained in benchmark treatment times for the Lidcombe Program (to be discussed shortly). The report was supplemented with a resource involving perspectives from participating clinicians about the rolling-group treatment model.<sup>45</sup>

## RANDOMISED CLINICAL EXPERIMENTS

According to the operational definition of a clinical trial presented during Lecture Five, a clinical trial involves evaluation of an entire treatment. The rationale given for such a criterion was that clinicians need information about the efficacy of an entire treatment to determine whether they might wish to use it. However, several reports have been published which have all the features of a randomised controlled trial according to this definition, with the exception that they are evaluations of parts of a treatment. These reports might be termed randomised clinical experiments.

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<sup>†</sup> Translational research is overviewed in Lecture Five.

## The Lidcombe Program

### *Sixteen weeks of Lidcombe Program compared to no treatment*

An experiment<sup>46</sup> with German children is sometimes cited as an independent replication of the Phase III Lidcombe Program trial,<sup>7</sup> and its title states that it is a clinical trial. However, the report involved only 16 weeks of treatment. Forty-six pre-school children, four of them girls, were randomised to receive either 16 weeks of the Lidcombe Program treatment or 16 weeks of no treatment. One child dropped out of the treatment group. After 16 weeks of treatment, the children in the Lidcombe Program had 1.6 %SS in everyday speaking situations and the control children 6.9 %SS. This result was statistically and clinically significant.

### *Twelve weeks of Lidcombe Program compared to no treatment*

With a similar design to the German study, an Australian experiment<sup>47</sup> randomised 29 pre-school children, four of them girls, to receive either 12 weeks of the Lidcombe Program treatment or 12 weeks of no treatment. Six children dropped out, leaving 10 in the treatment group and 13 in the control group. At 12 weeks post-randomisation, the children in the Lidcombe Program group had a mean 3.5 %SS during everyday conversations and the children in the control group had a mean 5.8 %SS. This result was statistically and clinically significant.

### *Twelve weeks of Lidcombe Program compared to RESTART-DCM*

As a preliminary study to the randomised trial discussed earlier,<sup>26</sup> a Dutch study<sup>48</sup> randomised pre-school children to a Lidcombe Program group and a RESTART-DCM group. Thirty children were randomised. Seven dropped out, leaving 11 children in the Lidcombe Program group and 12 in the RESTART-DCM group. Based on beyond-clinic recordings, results after 12 weeks of treatment were 3.7 %SS for the Lidcombe Program group and 3.1 %SS for RESTART-DCM. This result was clinically and statistically nonsignificant.

Interpreting their findings cautiously, and foreshadowing their later randomised trial, the authors concluded that “randomized controlled trials of LP versus DCM treatments are feasible” (p. 197).<sup>48</sup> They also correctly pointed out that further study of the matter is necessary with control groups in order to obtain fully interpretable results.

## **ANALYSES OF LIDCOMBE PROGRAM REPORTS THAT HAVE A CONTROL GROUP**

The ultimate endpoint of clinical trials research is a systematic review of meta-analysis for many randomised controlled trials (see Lecture Five). For clinical trials of stuttering, that is not yet possible, because not enough of them exist. However, the next best thing is an analysis of randomised clinical evidence for the Lidcombe Program that involves a no-treatment control group. That randomised clinical evidence includes randomised controlled trials,<sup>7,13</sup> and two randomised clinical experiments.<sup>46,47</sup> The mean post-randomisation period for those reports is 6.3 months.

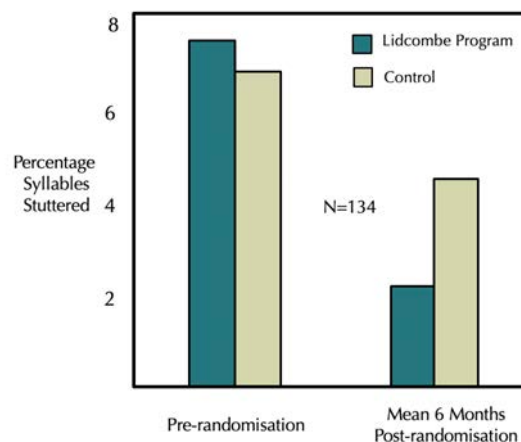
A Cochrane Systematic Review,<sup>49</sup> which has attracted some interest,<sup>50,51</sup> noted that the Lidcombe Program is unique with its demonstration of superiority over no-treatment controls, with analysis indicating a large effect size of 0.92 with a total of 151 children recruited:

This systematic review indicates that the Lidcombe Program may result in lower stuttering frequency and higher speech efficiency than a wait-list control group in children aged up to six years at post-test. However, these results should be interpreted with caution due to the very low and moderate certainty of the evidence and the high risk of bias identified in the included studies. (p. 2)<sup>49</sup>

Another analysis of these data<sup>52</sup> are shown in the figure. That analysis included 134 children in those reports for whom pre-treatment and post-randomisation data were available. At pre-randomisation the stuttering severity of the Lidcombe Program and control groups were about the same. There was some predictable improvement with the control children because of natural recovery. However, at a mean post-randomisation period of 6.3 months, the Lidcombe Program children did better than the control children.

The Lidcombe Program odds ratio was 7.5 for attaining below 1.0 %SS at 6.3 months post-randomisation. That means that, at 6.3 months post-randomisation, children who received the Lidcombe Program had 7.5 times greater odds of having “no stuttering” or “almost no stuttering” than children who did not receive the Lidcombe Program treatment. The 95% confidence interval was 2.7–20.9; there was an estimated 95% chance of the true odds ratio being between those two values.

It is necessary when interpreting this analysis to consider that, with a mean post-randomisation period of 6.3 months, not all the 134 children involved received the full treatment. So, the odds ratio for those children may have been greater had they received the full treatment. Therefore, it would be justifiable to conclude that the odds ratio is at least 7.5.



## DATA-BASED CASE STUDIES

For the present purposes, data-based case studies are reports published in peer-reviewed journals that are either retrospective—involving previously treated children—or reports that do not involve speech measures beyond the clinic, or which do not incorporate a clinically meaningful follow-up period. However, their conclusions about treatment outcome focus on speech measurement.

### The Lidcombe Program

Ten Swedish children were enrolled in a case study report of the Lidcombe Program.<sup>53</sup> Six of them completed the treatment and reduced stuttering, according to measures in the clinic, from a pre-treatment mean of 6.7 %SS to a post-treatment mean of 0.1 %SS at the end of Stage 2. The post-treatment assessment was “21 months or more after achieving fluency” (p. 251),<sup>53</sup> which presumably means 21 months after completing Stage 1.

Several other case studies have been reported for the Lidcombe Program with Chinese,<sup>54</sup> Japanese,<sup>55</sup> British,<sup>56</sup> Canadian,<sup>57</sup> Iranian,<sup>58</sup> and United States<sup>59</sup> children. The latter was a data-based follow up of 15 children 1–5 years after treatment. Based on video recordings of the children speaking in their homes, mean pre-treatment scores were 12.6 %SS and mean post-treatment scores were 0.5 %SS, which was a 96% reduction.

Several individual case studies of the Lidcombe Program have been reported with French Canadian children with early stuttering.<sup>60,61,62</sup> Those cases were consistent with a report of Malaysian children treated with the Lidcombe Program<sup>19</sup> that treatment in one language generalises to another language. A report with a Belgian child 3 years 4 months old<sup>63</sup> showed treatment success by telehealth. A study of five Iranian children (ages 4–6 years) using video telehealth<sup>58</sup> gave them only 15 clinical consultations and then (inexplicably) stopped their treatments. Regardless, one of the children appeared to approach criteria for entry to Stage 2 after 15 weeks of treatment.

### Palin Parent-Child Interaction Therapy

A retrospective file audit<sup>64</sup> of 55 children treated at the Michael Palin Centre in London involved 38 boys and 17 girls with a mean age 53 months at the start of the study. Percentage syllables stuttered was measured before treatment and 3, 6, and 12 months after the start of treatment. Measures were based on clinic video recordings “while the child described a series of “What’s wrong?” pictures with the SLPs [speech-language pathologists] in the clinic” (p. 1214).<sup>64</sup> The duration of the recordings was not specified. Additionally, children were measured with the KiddyCAT, which is a parent report measure of child attitude to communication (see Lecture Twelve).

From before treatment and 12 months after the start of treatment, %SS scores were, respectively, 6.7 and 2.3, which was a 66% reduction. This is consistent with the 64% reported for the 12 children in two clinical trials of the procedure,<sup>21,22</sup> discussed earlier. KiddyCAT scores reduced from 4.6 to 2.0, showing an improvement of the children's attitude to communication.

Another retrospective file audit involved three child stutterers with a comorbid diagnosis of autism spectrum disorder.<sup>65</sup> The children were ages 4 years 5 months, 6 six years 7 months, and 7 years 7 months. Dependent measures were %SS, the KiddyCAT, the OASES-S (see Lecture Four), and a measure of how stuttering impacts children and parents.<sup>66</sup> One of the children showed a clinically significant %SS reduction over 12 months, but no details were provided about how the measure was collected. KiddyCAT scores were reported for two children, with one improving and one worsening. There was some evidence (see Figure 6) of improved parent knowledge about stuttering and confidence with managing it.

### A family-focussed treatment approach based on a multifactorial model

There has been a case study report<sup>67</sup> of a treatment that is broadly similar to the treatments discussed previously based on multifactorial models: Palin Parent-Child Interaction Therapy and RESTART-DCM treatment. This treatment, developed at the Stuttering Center of Western Pennsylvania at the University of Pittsburgh, in part draws specifically on the Demands and Capacities Model. A treatment manual is available.<sup>68</sup>

Similar to the Palin Parent-Child Interaction Therapy treatment process, this family-focussed treatment approach "typically consists of six to eight sessions, 45 min in length, scheduled once per week or every other week" (p. 120).<sup>67</sup> It is also consistent with Palin Parent-Child Interaction Therapy that the treatment goal does not overtly specify no stuttering or nearly no stuttering as a treatment goal. Instead, the treatment

is designed to help young children who stutter (between the ages of 2 and 6) improve their speech fluency while simultaneously ensuring the development of healthy communication attitudes and effective communication skills. (p. 119)<sup>67</sup>

Also consistent with Palin Parent-Child Interaction Therapy, and RESTART-DCM treatment, is that "direct fluency shaping and stuttering modification" (p. 119)<sup>67</sup> procedures are implemented if needed. Another similarity is that the clinician works with parents to construct individual treatment plans according to need. The multifactorial "bucket analogy" (see the diagram in Lecture Three) is presented to parents during this process:

For example, if parents report that their schedule at home is busy and that they often feel rushed, and if they believe that this contributes to time pressures that affect the child's speech, then the parents and clinician may brainstorm ways of reducing these time pressures. The parents may then work to set aside a set period of time each day so the child can interact with the parents with less time pressure, or they may consider different scheduling options for the child's activities in order to allow for more one-on-one time. (p. 121)<sup>67</sup>

The specific "parent communication modifications" considered are as follows:

(a) use and modeling of an easier, more relaxed manner of speaking ... (b) use of increased pause time between speaker turns so as to reduce time pressures the child may feel when communicating; (c) reduction of demands to speak and increased time pressures often associated with "rapid-fire" questioning, if present; and (d) reflecting, rephrasing, and expanding on children's utterances to provide a positive communication model. (p. 123)<sup>67</sup>

The results of the report were fairly consistent with the two clinical trials of Palin Parent-Child Interaction Therapy,<sup>21,22</sup> indicating that six of 17 children "continued to stutter following completion of the parent-focused treatment" (p. 128).<sup>67</sup> Mean scores for "stuttered types of disfluencies per 100 words" (p. 126) were 16.4 before and 3.2 after the intervention. Parents scored "how often the child was able to speak *without [authors' italics] stuttering*" (pp. 126–127) with a 5-point scale: 5 = *always*,



4 = *almost always*, 3 = *sometimes*, 2 = *rarely*, and 1 = *never*. The post-treatment mean for three beyond-clinic speaking situations was around 2.8.

### A case study of several treatments

A report with six Iranian children with early stuttering<sup>69</sup> involved the Lidcombe Program, Palin Parent-Child Interaction Therapy, and a hybrid treatment combining the two. For several reasons, this report does not provide useful information for clinicians. No reason was given for combining the two treatments. Also, there are transparent problems in the report with treatment fidelity. For example, components of the Lidcombe Program are described which do not exist, such as “following children’s model in play,” “reducing parent’s speech speed,” and “observing turn taking in families” (p. 33).<sup>69</sup> Finally, all children received a 12-week combination of all three treatments, making the results uninterpretable.

## THERAPIST DRIFT

### The Lidcombe Program

#### *Therapist drift*

Departure from manualised procedures, or *therapist drift*<sup>70</sup> to use the correct term, is thought to be clinically undesirable. It may be a justifiable assumption that adherence to the Lidcombe Program Treatment Guide will produce optimal treatment results. However, currently there is little empirical support for that contention, but there are three reports that therapist drift with the Lidcombe Program does occur. These reports are now discussed.

#### *Therapist drift in a translational study*

During the translational study outlined previously,<sup>†</sup> the 31 community clinicians generally adhered to the Lidcombe Program Treatment Guide. However, around half of them varied from the prescribed 45–60 minute clinic sessions and used 30-minute sessions instead. Also, fortnightly clinic visits often occurred instead of the prescribed weekly visits, sometimes because of clinician scheduling and sometimes because of client failures to attend their clinic sessions. The mean number of days between clinic visits was 15.4, rather than the ideal seven days specified in the treatment guide. However, there was no evidence that these fidelity problems affected outcomes. The result that fortnightly clinic visits did not affect outcomes was consistent with another file audit of 134 North American pre-schoolers treated with the Lidcombe Program.<sup>71</sup>

The translational study also showed that around half of the 31 clinicians did not use the recommended procedure of having parents demonstrate verbal contingencies in the clinic each week. It was not possible to determine statistically whether this treatment fidelity problem affected outcome. Regardless, the authors concluded:

The fact that only half the SLPs [*speech-language pathologists*] in the present study routinely requested the parent to demonstrate treatment in the clinic, however, is concerning. Without observing parents demonstrating treatment, it is not possible for SLPs [*speech-language pathologists*] to confirm that parents are implementing the treatment safely and correctly, and that the child is responding positively. However, the study design did not allow exploration about why SLPs [*speech-language pathologists*] in the present study largely chose not to adhere to this important treatment procedure. (p. 601–602)<sup>†</sup>

#### *Therapist drift in two observation studies*

This important treatment fidelity issue—that parents may not be doing verbal contingencies the way the clinician has instructed them during the Lidcombe Program—was explored with three children during their treatment.<sup>72</sup> It was encouraging that the three parents presented more verbal contingencies for stutter-free speech than stuttered speech during practice sessions, as specified in the treatment guide. This was also the case with verbal contingencies during natural conversations, although more

contingencies for stuttering occurred in such situations.

The report had some sobering features. One parent continued with the treatment during practice sessions despite the child saying he did not like the activity. Another parent focused on the rules of the game being played rather than the child's speech during such treatment. All parents were observed to give incorrect verbal contingencies, such as praising stuttered speech, during treatment in practice sessions.

To assist clinicians with ensuring Lidcombe Program treatment fidelity, the report concerned<sup>72</sup> presents an empirically developed checklist of procedures for treatment during practice sessions, which focusses on the important issues of parent verbal contingencies. The paper contains two case histories of the checklist being used.

A larger study<sup>73</sup> involved 40 parent-child pairs during Stage 1 of the Lidcombe Program. The parents recorded the practice sessions they did each day and kept a diary of their use of verbal contingencies during natural conversations. There were some positive results. The mean duration of practice sessions was 12.7 minutes, and the median number of practice session per day was once per day. Those findings were consistent with the Lidcombe Program Treatment Guide. Most verbal contingencies during practice sessions were for stutter-free speech, as specified in the Treatment Guide; 91% were for stutter-free speech and 6.8% were for unambiguous stuttering, with only 2.7% of verbal contingencies being incorrectly applied.

However, according to parent diaries, the number of verbal contingencies during natural conversations was lower than expected: an average of 8.5 contingencies per day for stutter-free speech and 1.7 contingencies per day for stuttered speech. Also, an unexpected and puzzling association was found between the number of verbal contingencies for stuttering during natural conversations and the number of clinic visits to complete Stage 1. It was expected that more verbal contingencies would be associated with fewer clinic visits, but the opposite trend was reported.

#### *Therapist drift in survey studies*

A survey of 277 Australian speech-language pathologists<sup>74</sup> reported that around half of them said they departed from the procedures specified in the Lidcombe Program Treatment Guide. A common reason given for such departures was that Australian public health providers sometimes did not allow the full treatment to be given, allocating only treatment "blocks" of time to any one child, with the blocks of time not long enough for the treatment. (This is reminiscent of how British health care managers allocated treatment with Palin Parent-Child Interaction Therapy, as discussed during the previous lecture.)

Other problems documented in the survey were workplace service restrictions, including one report of children waiting up to 12 months for treatment. Other identified problems included allocating the requisite time in school settings that provided treatment services. For the study sample, 23% of clinicians were in schools. This is a particular problem in the United States where a public law states that all children who are disadvantaged because of disability must receive prompt remediation.<sup>75</sup> Consequently, United States clinicians with many children on their caseloads are by law not permitted to have a waiting list; all affected children must be treated promptly. In many such cases, children would not be able to receive a complete Lidcombe Program treatment as specified in the treatment guide.

Another study involved surveys of 215 Australian speech-language pathologists who had attended professional development training for childhood stuttering.<sup>76</sup> Interviews showed it was common for the speech-language pathologists to depart from procedures outlined in the Lidcombe Program Treatment Guide. Two key reasons for this were that weekly clinic appointments for 45–60 minutes were not viable in their workplaces, and that parents did not comply with daily severity ratings and treatment recommendations until the end of Stage 1. Common modifications to the treatment guide were not using parent severity ratings, not demonstrating procedures to parents, and not using practice sessions. The study also found, not surprisingly, that formal training in the Lidcombe Program was associated with higher clinician-reported success with the treatment.

## MECHANISMS OF ACTION

There are benefits to understanding the mechanisms of action for treatment effects: the “process by which the treatment’s essential ingredients induce change in the target of treatment.”<sup>77</sup> As argued in Lecture Three, if the mechanism of action of a treatment can be explained satisfactorily to clients and parents of children who stutter, compliance with that treatment may be enhanced because it is more likely to be perceived as credible, with an expectancy of positive outcome. Additionally, knowledge about mechanisms of action enables treatment developers to dispense with treatment components that do not contribute to treatment effects.<sup>78</sup>

### The Lidcombe Program

#### *The contribution of verbal contingencies*

The Lidcombe Program is based on laboratory studies showing that stuttering has operant-like properties because it responds to contingent stimulation (see Lecture One). The construction of the treatment around five parent verbal contingencies assumes that those contingencies are essential to the reported Lidcombe Program treatment effects.

An experiment<sup>79</sup> was designed to explore this assumption by randomising 34 parent-child pairs to two groups. The first group received the standard Lidcombe Program, and the second group received the Lidcombe Program without the verbal contingency *request self-correction*. The researchers measured the number of weeks and the number of clinic visits for the children to attain a 50% reduction of stuttering severity. They reported no significant differences between the groups. This result challenged the contribution of the verbal contingency *request self-correction* to the efficacy of the treatment, and it suggested the need for further research about the matter. This study supplements the study of parent treatment fidelity with verbal contingencies discussed earlier.<sup>73</sup>

The issue was further explored with a randomised controlled noninferiority trial.<sup>80</sup> The control arm was standard Lidcombe Program treatment, and in the experimental arm all the verbal contingencies were removed. Participants were 74 children aged between 3 years 0 months and 5 years 10 months, 37 of whom were randomised to each group. At 18 months follow-up, 31 children remained in the control group and 26 remained in the experimental group. The authors concluded that:

Findings of noninferiority were inconclusive for the primary outcome of stuttering severity, based on a margin of 1.0 percentage syllables stuttered. ... The inconclusive finding of noninferiority means it is possible that verbal contingencies make some contribution to the Lidcombe Program treatment effect. (p. 3419).

Together, the two studies raise an issue in need of resolution: how do verbal contingencies contribute to Lidcombe Program treatment effects?

#### *Child and parent language*

One suggestion<sup>81</sup> has been that children, or parents, might simplify their language production after the treatment. An initial report<sup>81</sup> with nine parent-child pairs found that not to be the case, with a range of parent and child language measures not changing from pre-treatment to post-treatment. Measures included speech rate, inter-speaker turn latency, mean length of utterance, developmental sentence scoring, number of different words, requests for clarification, and requests for information. No differences were found for the pre-treatment to post-treatment period. In fact, maternal speech rate increased after the treatment and parents decreased their rate of questioning. There was a slight suggestion, however, that the children did not meet full developmental language expectancies during the pre-treatment to post-treatment period.

Subsequently, the result of no pre-treatment to post-treatment language change was replicated with four children.<sup>57</sup> At post-treatment, the children increased their mean length of utterance, percentage of complex sentences, and number of different words. This result was replicated with another eight children,<sup>82</sup> showing no change of mean length of utterance, type-token ratio, and a phonological measure of percentage consonants correct.

A longer term study<sup>83</sup> was conducted on 11 children with early stuttering prior to Lidcombe Program treatment, at 9 months post-treatment and 18 months post-treatment. Measures were mean length of utterance, number of different words, and subordination index. During the period of study, there was no evidence associating Lidcombe Program treatment with restricted language development. The children showed no differences from the developmental trajectories of normative data for the three language measures.

#### *Acoustics*

There has been one attempt to find an acoustic explanation for the apparent efficacy of the Lidcombe Program;<sup>82</sup> perhaps children use a slightly different speech pattern post-treatment that controls stuttering. However, this report found no changes for vowel duration, intervocalic interval, voice onset time, or articulation rate.

#### *Inter-turn speaker latency*

In contributing to the search for Lidcombe Program mechanisms of action, researchers argued<sup>78</sup> that inter-turn speaker latency is a commonly cited feature of treatments based on multifactorial models that were overviewed in Lecture Six. This involves parents pausing briefly before responding to what a child says. They also drew attention to laboratory evidence—reviewed shortly—indicating that this variable may control stuttering. Consequently, they studied five speech-language pathologists while they were conversing with children and parents during Lidcombe Program clinical consultations. The speech-language pathologists used shorter inter-turn speaker latencies with the children than when speaking with the parents. The researchers concluded that it is possible for inter-turn speaker latency to be somehow vicariously associated with the Lidcombe Program treatment process, and that this possibility requires further research.

#### *Speculation about cortical plasticity*

It is thought to be reasonable speculation that the Lidcombe Program somehow rectifies issues with neural speech processing that are associated with stuttering (see Lecture Three). An efficacious treatment such as the Lidcombe Program occurs “while the brain features a remarkable capacity for plasticity” (p. 2).<sup>84</sup> Perhaps, then, the Lidcombe Program induces children to “adopt a compensatory neural growth pattern that successfully makes up for the deficient brain regions” (p. 77).<sup>85</sup> This idea has gained more recent support from other authors.<sup>86</sup> Another author has suggested a similar mechanism in terms of the malleable nature of the developing brain.<sup>87</sup> A report of dyslexic school-age children was consistent with this possibility, showing changes of grey matter volume after 8 weeks of therapy.<sup>88</sup>

This idea incorporates the well-known notion of “neuroplasticity” or “cortical plasticity.” Not only does the brain drive behaviour, but behaviour drives the brain. A review of the topic<sup>89</sup> presents converging lines of evidence that “suggest an active role for dynamic myelination in adult brain plasticity and indicate myelin plasticity may be an additional route by which experience can shape brain structure and function” (p. 86). For example, learning to juggle can change grey matter structure in areas that support visual learning.<sup>90</sup> Another study showed changes in the occipito-temporal cortex after only seven days of learning to juggle.<sup>91</sup>

If such mechanisms of action are involved with the Lidcombe Program, they could be verified experimentally, such as with brain scanning of experimental and control children before, during, and after a treatment period.

#### *Speculation about common factors*

The common factors model is that features shared by all therapeutic approaches contribute to their success more than anything that is unique to any approach.<sup>92,93</sup> This idea has been presented to explain the effects of the Lidcombe Program<sup>94</sup> along the lines of “the quality of the client-clinician relationship ... and ... the client and the clinician’s hope or expectation that change can and will occur” (p. 5). The authors argue that all historical and current treatment methods for stuttering owe their success to these factors more than anything else. This idea has some intuitive appeal but is

currently not supported by any research.

## Treatments based on multifactorial models

### *Parent language behaviour in general*

A review of the topic<sup>95</sup> and a subsequent empirical investigation<sup>96</sup> found no reason to associate unusual parent language behaviour with early childhood stuttering, or any reason to believe that changing parent language behaviour may be clinically useful with early childhood stuttering. However, that overarching statement requires some qualification considering recent reviews and research, which is now considered.

### *Parent question asking*

A study of parent questions in early childhood stuttering<sup>97</sup> was prefaced with an overview of the many recurring recommendations, based on the Demands and Capacities Model, that parents reduce the number of questions they ask when conversing with their children. The study involved 32 children with early stuttering and 32 controls, ages 28–50 months, and 15,782 utterances from a publicly available data bank of children's conversations with adults. For the children with early stuttering, responses to parent questions were significantly less likely to contain stuttering-like disfluencies than other utterances. In other words, based on the results of that study, clinical advice to emerge from the Demands and Capacities model is incorrect. The authors concluded that:

Given the current findings, some prior research, and the documented potential benefits in language development for adult question asking of children, we do not believe that clinicians need to recommend changes to typical question-asking behavior by caregivers of CWS [*children who stutter*]. (p. 2061)<sup>97</sup>

### *Parent reduced speech rate*

During RESTART-DCM treatment parents may reduce speech rate and increase their interturn speaker latency. With Palin Parent-Child Interaction Therapy those changes seem to occur often during treatment. Therefore, experimental evidence to verify the capacity of those variables to control early childhood stuttering is of interest. A review<sup>98</sup> of five laboratory experiments of parent reduced speech rate for children with early stuttering<sup>99</sup> concluded that stuttering reductions of around 50% were observed overall under such conditions. However, effects were not observed for every child studied. One of those experimental reports<sup>100</sup> concluded that study of the effects of extended, everyday parent use of such techniques is warranted.

### *Increased interturn speaker latency*

An experimental study of a 5-year-9-month old child who stuttered<sup>101</sup> involved increased interturn speaker latency for 15 sessions during a 7-week period at the family dinner table. The parents and the child's 10-year-old brother participated, with the children using wooden blocks to signal the need to have a conversational turn. Results suggested that the procedure was responsible for a 40–50% reduction of stuttering during the experimental conditions compared to baseline. A laboratory experiment<sup>102</sup> with three boys who stuttered involved three experimental sessions of "no interruption" for two of them during conversation with a researcher. For one boy, age 6 years 2 months, a stuttering reduction of around 50% was observed. No effect was observed for the other boy, age 5 years 6 months. A study of 27 children with early stuttering,<sup>103</sup> with a mean age of 4 years 0 months, showed that parents could be taught to slow their speech rate and increase interturn speaker latency. In the clinic, "stuttering-like disfluencies" (see Lecture Four) of the children decreased by 36%.

### *Parent recasts*

Parent recasts are a naturally occurring strategy of repeating a child utterance and adding to it. For example, if a child says "me want ice cream," a parent might say "do you want some of that ice cream?" Recasts may be simple or complex, they may add or modify a child's clause, and they may or may not correct the child's language. Parents of children with typical language and specific language impairments present recasts to their children around once per minute during conversations.<sup>104</sup>

In formulating a study of recasts as mechanisms of action in treatments based on multifactorial models,<sup>105</sup> researchers draw attention to two treatments that include them. They pointed out that RESTART-DCM treatment<sup>28</sup> does specify that parents use recasts, but only as a standard, helpful, and supportive parent strategy rather than as a response to stuttering. However, the researchers<sup>105</sup> indicated that another treatment based on a multifactorial model<sup>67</sup> does involve parent recasts as a response to stuttering.

The study used<sup>105</sup> a publicly available data bank of recorded conversations between child stutterers and adults. Participants were 43 children with a median age of 38 months. For the 43 children, there was a range of 0–25 utterances that contained stuttering, after which parents used a recast. For those utterances, compared to stuttered utterances that were not recast, there was significantly lower %SS scores: 4 %SS compared to 13 %SS. The effect might have occurred because children shortened their utterances after a parent recast. However, the experimenters showed that this probably was not the case, with no evidence of shortened child utterances after parent recasts. The researchers concluded that “naturally occurring conversational recasts of preschoolers’ stuttered utterances are fluency-facilitating” (p. 7). They also concluded that their study did not allow a conclusion that “recasts contingent on stuttered utterances at a given rate would also be fluency-facilitating” (p. 7).<sup>†</sup> However, they noted that their study showed that parents recast their children’s stuttered utterances at a low average rate of twice per 15 minutes.

### The Westmead Program

As noted in Lecture One, the immediate effects of rhythmic speech are striking, and have been known about for centuries. As outlined in Lecture Six, there is experimental evidence of its potential to control stuttering in 9–11-year-olds.<sup>106</sup> For adults, there is evidence of its associated acoustic effects.<sup>107</sup> With a study of four children, ages 8–11 years, who received the Westmead Program<sup>108</sup> the treatment, not surprisingly, induced post-treatment reduced variability of vowel duration. However, there was no corresponding change of speech rate or language use for the children. For one child, perceptual judgments by observers suggested some effects of rhythmicity post-treatment. A similar perceptual finding occurred for one of 19 children, ages 6–11 years, in a clinical trial of syllable-timed speech.<sup>109</sup> Regardless, there has been no report to verify these mechanisms of action for pre-school children who receive the Westmead Program. But it seems likely that the intended mechanism of action for the Westmead Program is, in fact, syllable-timed speech.

## TREATMENT SAFETY

### The Lidcombe Program

As noted in the previous lecture, a potential limitation of the Lidcombe Program is that it is possible for a parent to misuse the treatment and give verbal contingencies in a punitive and excessive manner. Indeed, during the early development of the Lidcombe Program, concerns were raised that the treatment might send an overall negative message to children that would affect their self-esteem and establish unhelpful cognitions.<sup>110,111,112</sup>

In response to those concerns, the Lidcombe Program developers verified for eight children with early stuttering that the treatment is psychologically safe.<sup>113</sup> Measures with the Child Behavior Checklist<sup>114</sup> showed no behavioural indications of any changes with the children pre-treatment to post-treatment that might suggest anxiety, aggression, withdrawal, or depression being associated with the treatment. Additionally, the Attachment Q-Set,<sup>115</sup> which measures the strength of bond between parent and child, showed that there were no changes after treatment. In fact, if anything, attachment appeared to improve. These results were confirmed by the randomised trial discussed earlier,<sup>26</sup> which reported some suggestion of post-treatment improvement for Child Behavior Checklist and KiddyCat scores

<sup>†</sup> It is conceivable that, in addition to increased interturn speaker latency as discussed earlier, parent recasting of stuttered utterances is a vicarious mechanism of action involved in the Lidcombe program.

after Lidcombe Program treatment. A clinical trial of the Lidcombe Program extended findings of psychological benefit to 7–12-year-olds,<sup>116</sup> with post-treatment improvements in anxiety, communication attitude, and impact of stuttering (see Lecture Nine).

Naturally, though, findings about the safety of the Lidcombe Program pertain to clinically appropriate management of any threats to safety. If there is any chance that a parent will use verbal contingencies in a punitive manner, the clinician needs to deal with that situation without delay.

### Treatments based on multifactorial models\*

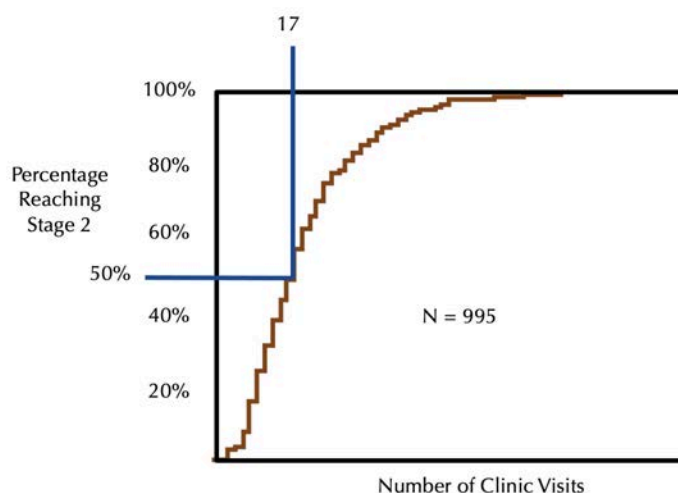
As with the Lidcombe Program, there is a basic issue about the safety of these treatments that needs to be dealt with. Treatments based on multifactorial models change features of everyday childhood life that appear essential to healthy development. Active participation in conversation<sup>117,118</sup> and sustained interaction with adults<sup>119,120</sup> is known to be fundamental to early linguistic development. A review of three decades of literature<sup>121</sup> presented four critical aspects of healthy oral language childhood development: “family dynamics, ... interaction with parents, immediate social environment, and encouragement given to the child in the first years of life” (p. 350). As yet, there has been no research directed at the effects of changing these features of early childhood life during treatments based on multifactorial models, but obviously it is required.

### HOW LONG DOES TREATMENT TAKE?†

#### The Lidcombe Program

There is a sufficiently comprehensive data set to give an indication of how many Stage 1 clinic appointments may be required with the Lidcombe Program; in other words, how many clinic appointments are required to attain “no stuttering” or “nearly no stuttering.” More than a thousand children have been participants in Lidcombe Program clinical research, and the following figure\* contains information about treatment time for children, based on reports reviewed earlier: five file audits, seven clinical trials, two prospective follow-ups, two translational studies, and one prospective observation study. Those studies, involving a total of 995 children, reported a median number of clinic appointments to attain Stage 2 criteria. The average reported median clinic visits across those studies was 17.‡ The range of median clinic appointments reported in those studies to attain Stage 2 is 10–30.

The graph of these data in the figure is



\* Thanks to Juliet Imeson for guidance with material in this section.

† Thanks to Michelle Donaghy for guidance with material in this section.

\* Adapted and reproduced with permission: Jones, M., et al. (2000). Treating stuttering in young children: Predicting treatment time in the Lidcombe Program. *Journal of Speech, Language, and Hearing Research*, 43, 1440–1450. © 2000 American Speech-Language-Hearing Association.

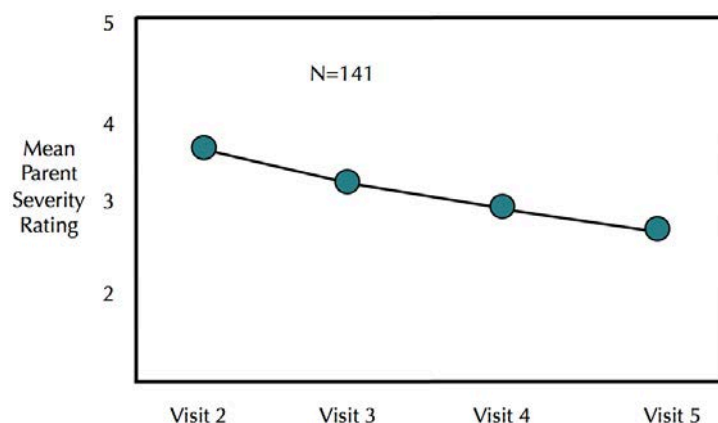
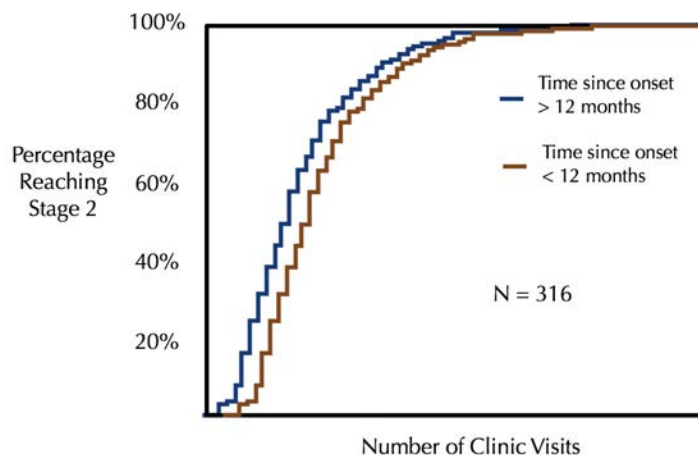
‡ Two of these publications contain around 40% of the cohort (N=316) who were treated during 1989–2001. At that time, the Lidcombe Program criteria for progression to Stage 2 needed to be attained for 1 week only, not the three consecutive weeks specified in the current treatment guide. These reports contain a median of 11 clinic visits, compared to 16 for the remaining studies. Consequently, the treatment times in these reports were adjusted by adding five more clinic visits to their median values, bringing them into line with the subsequent studies.

known as a recovery plot, and it shows the proportion of children to attain Stage 2 and the number of clinic appointments to do so. With the median number of clinic appointments being 17, half of those 995 children required fewer than 17 clinic appointments for Stage 1, and half required more than 17 clinic appointments. The estimated 90<sup>th</sup> percentile for the recovery plot is 28 clinic appointments. In other words, 90% of cases will have attained Stage 2 by 28 clinic appointments.

An important note here is that these data describe trends in large groups of clinical children, as do the results of the clinical trials described earlier. As such, care is needed in evidence-based reasoning to form a judgement about the extent to which these data apply to any individual clinical child. One consideration will be comorbid diagnoses and case features. Two reports,<sup>122,123</sup> for example, describe nine children—3% of the caseload—who dropped out of Lidcombe Program treatment because of comorbid speech and language problems, challenging behaviours, and complex family problems. Indeed, little is currently known about treating children with speech disorders that are comorbid with stuttering.<sup>124</sup>

Additionally, the treatment time data just described pertain to a population of clinicians, and care is needed about applying them to an individual clinician. Treatment times for individual clinicians will vary according to the nature of their caseloads and their clinical experience and training.

The next figure\* is the mean parent SR for a report of 141 cases<sup>125</sup> showing an average reduction of around one third during the first five clinic appointments (4 weeks of treatment).



## HOW DOES A TREATMENT DELAY AFFECT THE TREATMENT PROCESS?

### The Lidcombe Program

The next figure,<sup>†</sup> shows the results for 316 children in the studies mentioned earlier.<sup>122,123</sup> The recovery plot on the left shows the children who had been stuttering for more than 12 months, and the recovery plot on the right shows the children who had been stuttering for less than 12 months. The children who had been stuttering for less than 12 months have the same shape of recovery plot as the others,

\* Adapted and reproduced with permission: Onslow, M., et al. (2002). Beyond-clinic speech measures during the Lidcombe Program of early stuttering intervention. *ACQuiring Knowledge in Speech, Language and Hearing*, 4, 82–85. © 2002 Speech Pathology Australia.

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however it is moved to the right to a statistically significant extent. This means that both groups of children, overall, responded in the same way to the treatment, but the children who had been stuttering for less than 12 months required a few more clinic appointments to reach Stage 2.

So, in terms of important clinical significance, it appears that delaying the Lidcombe Program for up to a year after stuttering onset is unlikely to jeopardise a child's responsiveness in terms of time taken to reach Stage 2. This research result is intuitive, considering, as discussed during the previous lecture, that the Lidcombe Program involves a cognitive requirement for children to understand the treatment process. It appears from these results that children who have been stuttering a little longer, and consequently are a little older and more cognitively developed, are a little more responsive in terms of time taken to reach Stage 2. In short, there are empirical and logical grounds to suggest that the Lidcombe Program is not optimally suitable for very young children who have begun to stutter.

## DO CASE VARIABLES AFFECT THE TREATMENT PROCESS?

### The Lidcombe Program

#### *Pre-treatment stuttering severity*

Six studies have used a statistical technique called logistical regression to predict treatment time with the Lidcombe Program.<sup>4,42,59,71,122</sup> These analyses show that pre-treatment stuttering severity accounts for around 20% of the number of Stage 1 clinic visits required. This finding is intuitive; if there is more stuttering it takes longer to control it. The prospective observation study of parents doing the Lidcombe Program discussed earlier<sup>73</sup> reported the same effect with a strong association. The study of the Lidcombe Program in a student clinic<sup>126</sup> also reported that effect. In a study of a clinical caseload,<sup>127</sup> the 10 children who took the longest to complete Stage 1 had more severe stuttering than 10 children who completed Stage 1 in the shortest time.

Another clinically useful perspective on this matter was presented in the Lidcombe Program translation report with community clinicians.<sup>†</sup> There was a 17% increase of Stage 1 clinic visits for every one SR scale value pre-treatment. So that could make quite a difference for a child with SR 4 compared to a child with SR 8. Such difference could prompt a clinical decision to begin intervention earlier with a more severe child so that treatment is completed before the school years.

#### *Phonological and language development*

There is logistical regression evidence in one clinical trial<sup>4</sup> that phonological development does not predict treatment time. However, this report found that, together with pre-treatment stuttering severity, receptive language scores and language development (Mean Length of Utterance), predicted 34% of the variance of clinic visits needed for Stage 1.

The result is a little difficult to understand, because better language development was associated with shorter treatment time, but higher receptive language scores were associated with longer treatment time. The former result seems intuitive, but it is not at all clear why better receptive language would be associated with longer treatment time. Without replication of the result, it is possible that the latter, unintuitive result is a Type II statistical error, where a finding is reported when in fact it is not true.

#### *Siblings treated previously*

The study of the clinical caseload mentioned previously<sup>127</sup> reported a novel, but intuitive finding. One variable that characterised the children in the short treatment time group was that a sibling was more likely to have been treated with the Lidcombe Program previously.

#### *Predicting treatment outcome*

All reports discussed so far have dealt with predicting treatment *time* for the Lidcombe Program, but there is one report that predicted treatment *outcome*.<sup>128</sup> For a cohort of 277 children who received Lidcombe Program treatment, 32 variables were used to predict short-term and medium-term treatment outcome. Outcomes at 6–9 months and 12–18 months after the start of treatment were

measured with parent report of stuttering severity. The study also explored predictors of whether parents would drop out of treatment. The 32 predictor variables spanned domains of demographics, stuttering severity, child speech and language, and child and parent psychometrics.

Results were that better language skills and “easy” childhood temperament (see Lecture Eleven) were statistically associated with better treatment outcomes. However, those results, albeit intuitively correct and statistically significant, were not clinically significant. Those variables only accounted for a minute portion of the variance of treatment outcome. No predictors of treatment dropout were found except—intriguingly—that parents who failed a personality screening relating to their impulsivity were 3.5 times more likely to drop out of treatment than parents who did not. The authors cautioned that the latter finding requires replication with a full personality assessment before it can be given credence.

## PARENT EXPERIENCES

### The Lidcombe Program

#### *The importance of this topic*

There have been three studies of how parents experience the Lidcombe Program.<sup>129,130,131</sup> These reports provide useful information to forewarn clinicians about both positive and negative features of the treatment that parents might encounter. Clinicians can be forewarned about potential adverse parent experiences. This material is supplemented with an interview report<sup>132</sup> of a parent and child 7 years after completing the Lidcombe Program.

One report<sup>129</sup> surveyed 35 parents whose children had recently completed Stage 1 of the treatment. Results showed the Lidcombe Program to be a generally positive experience for parents. Some parents reported lacking confidence to do it, but valued clinician support with overcoming that feeling. Some parents reported a sense of empowerment with doing the treatment. Some parents reported difficulty doing the treatment, mainly with finding the time each day for it. Most parents reported a positive response from children to the treatment, but some children did not like being interrupted by parent comments. There were no parent reports of adverse reactions, such as reduced talkativeness, and “in fact, many parents reported their child was more confident and more talkative as a result of the treatment” (p. 422).

Another report<sup>130</sup> found three typical paths (outlined below) through the treatment based on interviews with 14 parents of children who were being treated. Six of the parents were interviewed on two occasions. All but one interview was with mothers.

#### *Path One: Straightforward parent experiences*

The first path reported was a straightforward one, with parents enthusiastic and innovative. They easily incorporated the treatment into their lifestyles in an enjoyable manner and were able to do some independent problem solving instead of relying overly on the clinician. These families attained a quick and steady therapeutic response, with the child assuming some responsibility for the treatment.

#### *Path Two: Straightforward parent experiences then problems*

The second treatment path reported was a straightforward one initially, with problems subsequently encountered. Parent guilt about not being able to commit properly to the treatment, and guilt about stuttering itself, began to emerge. These parents became needy of support, the visits to the clinic became a burden for them, and they found the treatment difficult to sustain. The children of these parents began to be unresponsive, and even irritated, by parent verbal contingencies.

#### *Path Three: Problems from the outset*

The third treatment path through the Lidcombe Program involved encountering problems from the outset. These cases were in the minority though. Such parents had trouble doing the verbal contingencies and were not particularly adept at leading the child to do the treatment. These parents

would doubt their capacity to do the treatment, and they focussed on problems they were having rather than how to solve them. These parents, more than the others, talked about “their anxieties, feelings of inadequacy, guilt and distress” (p. 24).<sup>130</sup> The beliefs of such parents about stuttering and parenting were not a good fit with the Lidcombe Program. It seems clear that if the Lidcombe Program continues with sporadic or little progress, such parents can become distressed.

#### *Treatment implementation problems*

Another report<sup>131</sup> involved 16 mothers who were each interviewed nine times during treatment during a 6-month period. The key findings of the report dealt with treatment implementation, perception of the treatment, and parent emotions.

The first finding dealt with obstacles implementing the treatment. Those obstacles can be summarised as

- (1) Problems finding time to do the treatment
- (2) Forgetting to do the treatment
- (3) Problems managing siblings throughout the treatment.

Fourteen of the 16 mothers had more than one child and so issue (3) was prominent. Despite these implementation problems, several benefits were reported, including an increase of quality time with the children, along with an improved bond between them, and increased knowledge about stuttering. Improved parenting skills was also a feature of the treatment, which is not surprising because it is a behavioural treatment that places some demands on children.

#### *Parent perceptions*

Another emerging theme was about treatment credibility and expectancy. As discussed during Lecture Three, these are important issues related to treatment compliance. Mothers’ perception of the treatment could be too ambitious, expecting it all to be over in a few weeks. And there was also an issue of just not expecting it to work:

I wouldn’t hesitate for anyone to try it because I didn’t think it was going to work. I didn’t think that saying smooth and bumpy talking was really going to make such a difference and it did. (p. 76)<sup>131</sup>

Parents could also be taken by surprise by not expecting to have to do the treatment themselves and anticipating that the clinician would do the job for them. Parents offered suggestions about how the treatment could improve and proffered the need for “bigger picture” information about the treatment, more treatment documentation, and a support group.

Another aspect of this theme was the children’s reaction to the treatment. It was commonly reported that they were becoming aware of when they were stuttering and, therefore, self-evaluated stuttering. Eventually, they became more self-confident and lost some of their shyness. Direct signs of children enjoying the treatment were reminding parents to do treatment, and clear signs of enjoying praise for stutter-free speech. There were, however, two emerging topics suggesting negative reactions to the treatment. These were negative reaction to verbal contingencies and suggestions that the child had done something bad by stuttering. Interestingly, some children seemed to react negatively to the word “smooth” and reacted better when parents substituted something different like “great talking.”

#### *Parent emotions*

Parents were reported to experience nine emotions during treatment.<sup>130</sup> The five most common were judged as those emotions that appeared most strongly and were most often mentioned, and how many mothers reported them. The first was “empowerment and responsibility,” with parents realising it was up to them alone to incorporate the treatment into their lives. The accompanying responsibility could lead to anxiety and pressure to perform well with the treatment. Indeed, “anxiety” was a strong theme emotion. Parents could be concerned about doing the treatment properly and by a fear that their children were being teased and bullied at school and worried that their children would still be stuttering when they went to school.

“Parent guilt” occurred for many reasons, such as a belief it was they who caused the stuttering, guilt that they were not doing the treatment correctly, and guilt about not finding time to do it during the day. Eight mothers reported distress related to stuttering severity and the experience of having to watch the child stutter during treatment. The final parent emotion was referred to as a “cycle of confidence.” Mothers’ confidence followed the ups and downs of their children’s stuttering and their success in implementing the treatment. This cycle of confidence was reported throughout the entire 6 months of the study.

## **THE EARLY CHILDHOOD STUTTERING INTERVENTION EVIDENCE BASE: SUMMARY AND CONCLUSIONS**

### **The Lidcombe Program**

Eleven independent reviews consistently report that the evidence base for the Lidcombe Program is the most comprehensive available among early childhood stuttering treatments.<sup>49,133</sup> This evidence base includes nonrandomised clinical trials, nine randomised trials, randomised clinical experiments, case studies, treatment process reports, and qualitative studies of parent experiences. There are some compelling advantages in using video telehealth with the Lidcombe Program. Research publications continue to emerge, as described earlier during this lecture. A survey that included 124 Australian child stutterers, with a mean age of 11 years,<sup>134</sup> reported that 50% of them had received the Lidcombe Program.

Treatment fidelity, or implementation fidelity, refers to whether a treatment is administered as intended, and is an important consideration with treatment translation in general,<sup>135,136</sup> and the issue is thought to be important with stuttering treatment,<sup>137,138</sup> and particularly so with randomised trials that compare stuttering treatments for early childhood stuttering.<sup>139</sup> A review of implementation fidelity<sup>140</sup> for behavioural, parent-implemented treatments for pre-schoolers included a majority of studies of the Lidcombe Program. No study reported ideal components of implementation fidelity, leading the authors to conclude that “rigorous measurement and reporting of [*fidelity implementation*] in future intervention studies is required in order to better inform evidence-based practices for interventions with [*children who stutter*]” (p. 1).

That said, a strength of the Lidcombe Program evidence base is that it involves direct assessment of treatment effect size from randomised control trials and randomised controlled experiments that compared the treatment with a no-treatment control group. Another strength of the evidence base is that it contains replicated findings that are independent of the original Lidcombe Program developers, most notably the randomised trial comparing the Lidcombe Program with RESTART-DCM.<sup>26</sup> That said, there has been no replication of the randomised controlled trial of the Lidcombe Program<sup>7</sup> showing an effect greater than natural recovery in a no-treatment control group.

For more than two decades, research has sought to establish the mechanism or mechanisms that underlie the Lidcombe Program treatment effects. This research continues today, but at present, the mechanism, or mechanisms, underlying the Lidcombe Program remain unclear. Their discovery may eventually lead to the development of a treatment with much different characteristics to the current structure of the Lidcombe Program.

### **Treatments based on multifactorial models**

A strong feature of the evidence base for this style of treatment is that it includes the largest randomised clinical trial of a stuttering treatment reported to date,<sup>26</sup> although positive clinical trial results for this style of treatment await replication. Replication is a substantive issue here, considering that the non-randomised Phase I trials of Palin Parent-Child Interaction therapy did not produce convincing evidence of a treatment effect. Those trials showed a quarter of the children with post-treatment stuttering severity below 1.5 %SS, compared with three-quarters of the children in the randomised trial. Replication of the results of clinical trials of treatments based on multifactorial models is confounded by the fact that two of its prominent treatment variations—RESTART-DCM and

Palin Parent-Child Interaction Therapy—have clinically dissimilar treatment goals. It has been argued that a limitation of the evidence base for treatments based on multifactorial models is that, in contrast to the Lidcombe Program, it contains no direct estimate of effect size for the treatment derived from comparison with a no-treatment control group.<sup>39</sup> But as noted earlier, the RESTART trial suggests that effect sizes for RESTART-DCM, in terms of percentage stuttering reduction, are no different from those obtained with the Lidcombe Program.

### The Westmead Program

This treatment is at the early stages of its development. Data available are nonrandomised Phase I and Phase II trials, and one randomised controlled trial comparing it to the Lidcombe Program. However, the latter trial was severely compromised by high drop-out rates. To be as compelling as the evidence that is available for the Lidcombe Program and RESTART-DCM treatment, evidence from further randomised trials will be required.

### Discussions about the early stuttering intervention base

A report of a 2022 symposium<sup>141</sup> focused on the treatments for which there are randomised controlled trials. A clinical situation was considered where parents of a 3-year-old asked a clinician what outcome might be expected from early stuttering treatment. Three clinicians provided videos of themselves responding to such a clinical situation, and their responses generated elaborations and discussion about their evidence-based clinical reasoning.

One communication to parents focused on stuttering reduction. That approach involved “zero stuttering or very near zero stuttering” (p. 5), and to “remain stutter-free in the long term” (p. 6) as a reasonable goal based on available evidence. Caveats to that approach were that the clinician requires sufficient skill with the treatment method chosen, and that treatment involves “support and guidance” (p. 8) for children and parents. A more cautious and circumspect approach involved methodological reservations about the available evidence base. Those reservations extended even to whether stuttering severity is a suitable primary outcome for clinical trials of early intervention.

Such reservations emerged also in a discussion between two researchers and two SLPs discussed the issue of whether early intervention can stop stuttering development.<sup>142</sup> There was agreement that the evidence base for stuttering intervention has limitations and should be interpreted cautiously. That discussion also highlighted disparate views about the best outcome of stuttering intervention. One view was that reducing stuttering severity is an appropriate focus of early intervention, and another view focused on “ease of communication, anticipation of stuttering, and covert stuttering” (p. 1).

As noted during Lecture Five, evidence-based reasoning is not a rulebook that prescribes clinical practices, but is an opportunity for “the conscientious, explicit and judicious use of current best evidence in making decisions about the care of individual patients” (p. 71).<sup>143</sup> That conscientious and judicious reasoning process often will lead clinicians to different interpretations of clinical research, and consequently how that research should apply to clinical practice. The 2022 Symposium is a prime example of such different endpoints of evidence-based clinical reasoning.

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## LECTURE EIGHT: EVIDENCE-BASED SPEECH RESTRUCTURING TREATMENTS FOR PERSISTENT STUTTERING

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### SPEECH RESTRUCTURING TREATMENT

#### Background

##### *Overview of speech restructuring*

Speech restructuring refers to the use of a novel speech pattern to reduce stuttering or eliminate stuttering while sounding as natural as possible.<sup>1</sup> This technique is sometimes referred to as *fluency shaping*. During speech restructuring, clients learn to speak initially with a slow, drawling speech pattern that is stutter-free. The speech pattern is then shaped toward stutter-free speech that is as natural sounding as possible. Surveys of those who have sought treatment for stuttering<sup>2,3,4</sup> confirm that this approach is desirable for many of them.

##### *Terminology*

There are many variants of this clinical technique currently in use, referred to with many different terms: *prolonged speech*, *smooth speech*, *easy speech*, and *precision fluency shaping*. They include target speech behaviours taught to clients such as *reduced speech rate*, *extended vowel production*, *light articulatory contacts*, *gradual onset of vocalisation* (also known as *gentle onsets*), and *continuous breath flow during speech*.

##### *The mechanism of speech restructuring*

All these speech pattern techniques sound quite similar, and it is quite likely that they all work in essentially the same way to control stuttering. The target speech behaviours of the treatment have been associated with post-treatment acoustic changes such as reduced articulation rate, reduced duration of phonation intervals, reduced variability of vowel duration, increased voice onset time, vowel duration, and intervocalic interval.<sup>5</sup> However, no specific underlying acoustic mechanism has been found that might explain how the treatment functions. As discussed during Lecture Three, it is a theoretically tenable idea that stuttering moments reflect atypical neural speech processing. If this is so, it is plausible that these speech patterns somehow, in a manner currently unclear, offset that issue.

Indeed, there is evidence that a change to atypical neural processing occurs after speech restructuring treatments.<sup>6</sup> Some speculation has been offered about how this might involve neural speech networks being reorganised in response to speech restructuring.<sup>7</sup>

First, brain structures that are implicated in stuttering can be mobilized, indicating a neural response to the intervention. ... Second, brain activity and connections can be normalized. Fluency-shaping, involving slow speech, gentle vocalizations, and lighter movements, can even out brain activity differences between people who stutter and those who do not. ... Third, functionally maladaptive structures can become uncoupled, suggesting the adult brain's ability to discard ineffective pathways. ... Finally, intact speech motor learning related structures can become more strongly integrated, underscoring the adult brain's capacity to utilize functional connections. (pp. 9–10).

##### *A variant: Stuttering modification*

The term *stuttering modification* (sometimes *stutter more fluently*) is related to speech restructuring. Although it appears to be used less commonly these days, it refers to an alternative approach to controlling stuttering with a novel speech pattern. Charles Van Riper was an extremely influential clinician, largely because he developed the "stutter more fluently" technique.<sup>8</sup> The technique is not intended to impose an overarching speech pattern to control stuttering. Instead, the technique provides a way for clients to stop or reduce struggle with individual stuttering moments. Van Riper and

a colleague have described these techniques in detail.<sup>9</sup> Terms for the components of this technique are *cancellations*, *preparatory sets*, and *pull-outs*. An overview of these techniques and the history of their development are described in a reference text (pp. 429–431).<sup>10</sup> However, as noted there,

“stuttering modification” ... is a popular therapy in many places, although large-scale data verifying its effectiveness are still relatively sparse more than half a century after initial reports of its use. (p. 431)

Van Riper did report treatment data in 1958,<sup>11</sup> using methods that were much different to the standards of treatment outcome research today. However, the authors of the reference text note that:

... about half the AWS [*adults who stutter*] that he treated had very fluent or essentially typical speech, were largely free from fear and avoidance, and had improved social interaction five years after treatment. Many more were apparently much improved. (pp. 430–431)<sup>10</sup>

In the 21<sup>st</sup> Century there have been two reports of adult treatment that have incorporated Van Riper’s techniques,<sup>12,13</sup> although neither report conformed to the modern standards for a clinical trial of stuttering treatment provided in Lecture Five. The first report had all the features of a clinical trial except that speech measures were made in the clinic at 6 months post-treatment, and the second report was a file audit. One study<sup>14</sup> involved a single 135-syllable audio recording of a monologue spoken with pull-outs, and another spoken with preparatory sets. The perceptions of 62 university students were unfavourable compared to unmodified stuttered speech. However, this brief assessment of a single speaker does not seem to be a fair evaluation of such a multidimensional treatment method. There has been a clinical trial of Van Riper’s techniques adapted for children 7–12 years old,<sup>15</sup> which is discussed in Lecture Nine.

#### *Another variant: Voluntary stuttering*

This is another commonly recommended procedure that is related to stuttering modification, which, again, has limited research about its clinical value. It is also referred to as *negative practice*, *pseudostuttering*, and *bouncing*. Although it is a stuttering modification behaviour, it is designed “to reduce fear, anxiety, and/or negative emotions associated with stuttering” (p. 290).<sup>16</sup> As such, it is classifiable within anxiety management strategies known as *behavioural experiments*, which will be discussed during Lecture Twelve. The report just mentioned<sup>16</sup> surveyed 206 participants who had knowledge of the technique, and reported that around half reported that it assisted with their fear of stuttering, and around a third reported that it made them feel more confident with their speech. It was clinically important, though, that the report noted that around two-thirds of the clients reported discomfort using the technique and that “when they first used voluntary stuttering it was too emotionally difficult for them to use in everyday situations” (p. 295).<sup>16</sup>

## **A brief history of speech restructuring**

### *A long time ago ...*

The history of speech restructuring has been described in detail.<sup>17</sup> Lecture One outlined how Satyrus seems to have used rhythmic speech during the Third Century BC to help Demosthenes with stuttering. This appears to be the first recorded use of a novel speech pattern to control stuttering. It is also generally believed that Satyrus recommended that Demosthenes speak above the roar of an ocean. It is completely possible that this was another use of speech restructuring to assist with stuttering; speaking with increased volume may have induced a novel speech pattern.

### *Twenty centuries later ...*

This brief historical overview jumps some 20 centuries to 1724, when an American minister of religion, Cotton Mather, published the following in a medical treatise, which describes a technique that has obvious conceptual similarity to modern speech restructuring:

While you go to snatch at Words, and are too quick at bringing of them out, you’ll be stop’d a thousand Times in a Day. But first use yourself to a very

deliberate Way of Speaking: a Drawling that shall be little short of Singing. Even this drawling will be better than Stammering; especially if what you speak, be well worth our waiting for.<sup>18</sup> (p. 460)

The use of speech restructuring variants occurred many times during subsequent centuries, and a full historical account is given in a 1984 text.<sup>19</sup> However, the present brief account jumps to 1951, when the effects of delayed auditory feedback—described during Lecture One—were reported. The oddities of speech under the influence of delayed auditory feedback were referred to initially as “artificial stutter.”<sup>20</sup> This began a long period of research about the effects of delayed auditory feedback on stuttering, which eventually did not lead to any real understanding about the nature and cause of stuttering.

*The 1960s onwards ...*

Although the discovery of the effects of delayed auditory feedback did not yield any theoretical insights, its clinical impact was extensive. During the early 1960s Israel Goldiamond discovered that those who stutter could overcome the effects of delayed auditory feedback by using a slow and drawling speech pattern. As it happened, that speech pattern could keep stuttering in check. He coined the term *prolonged speech* for this speech pattern, published a report about its clinical value in 1965,<sup>21</sup> and a revolution began with treatment for persistent stuttering. Goldiamond’s technique was to establish a novel and slow speech pattern to control stuttering, using a delayed auditory feedback device. The next steps of treatment were that the delay was systematically reduced, and speech rate was systematically increased.

In the reference text mentioned earlier<sup>19</sup> there is a detailed historical account of this style of treatment spreading through the Western World, and probably beyond. The use of delayed auditory feedback was soon found to be unnecessary and was replaced with recorded models of the requisite speech pattern and clinical instruction. There have been more clinical trials of speech restructuring treatment, with more participants, and with more independent replications, than for any other stuttering treatment. By the end of the 1970s there was sufficient research for a meta-analysis of the efficacy of speech restructuring treatments, and a conclusion that the method was more efficacious than any other.<sup>22</sup> According to a 2020 review, that conclusion still holds,<sup>23</sup> with it being the most promising way for adults to control their stuttering, should they wish to do so. A survey that included 625 Australian adult stutterers<sup>24</sup> reported that around a quarter of them had received a speech restructuring treatment.

## PROGRAMMED INSTRUCTION

### A technique for behavioural control

Often, speech restructuring treatments reported worldwide incorporate programmed instruction, which is a technique invented by the behaviourist B. F. Skinner. Its principles and their application to speech-language pathology have been outlined in detail.<sup>25</sup> It is a technique for learning behavioural control—of stuttering in this instance—with small increments arranged in a hierarchy of what is presumed to be easier to more difficult. Clients learn to master the hierarchical increments in small steps within a pre-determined sequence.

### Performance contingent progression

Some clients will take longer than others for any given programmed instruction sequence, and one reason is that progress through the incremental steps is performance contingent. In other words, there is a criterion, or several criteria, for completing each of the incremental steps. For example, one of the criteria for completing an incremental step might be no stuttering. In which case, if a stuttering moment occurs while a client is attempting to complete a step in the programmed instruction hierarchy, the client returns to the start of the sequence.

### A fundamental assumption

Programmed speech restructuring contains a clinical assumption that each step in the hierarchy is



more difficult than the previous one for the client to achieve. Therefore, it is also assumed that moving through the hierarchy is a productive way to learn, and that success at one step depends on success at the previous step. Those assumptions may well be true, but for stuttering treatment they are not substantiated by any research.

### Models of programmed instruction

Most speech restructuring clinical trials have involved programmed instruction. Commonly, intensive treatment formats are used. These raise health economics issues about efficient use of clinical resources. For example, there has been an observation<sup>26</sup> that the results of a 5-day, non-residential intensive treatment<sup>27</sup> appear similar to the results of a 3-week residential treatment.<sup>28</sup>

In fact, intensive speech restructuring treatment does not seem essential to a positive treatment outcome. A clinical trial<sup>29</sup> suggested that 10 one-hour sessions of individual treatment produced equivalent results to a treatment version involving an intensive treatment day. And there has been a clinical trial<sup>30</sup> showing that 16 two-hour sessions over four consecutive days produced equivalent results to two 2-hour sessions for 8 weeks.

With many programmed instruction stuttering treatments, the hierarchical sequence involves speech rate increments, commonly syllables per minute (SPM). The target speech rate increases with a sequence of six steps from extremely slow, such as 50 SPM, to a target speech rate somewhere near typical rates, or appreciably slower, such as 200 SPM. It is not realistic to require clients to speak with their speech rate exactly at specified SPM values, so they are given some leeway, such as plus or minus 20 SPM. There are several speaking tasks within each step, usually from five to 10. The speaking tasks might involve monologues, or conversation with a clinician.

### Instatement and transfer

The part of treatment just described is often referred to as *instatement*, and sometimes as *establishment*. Subsequently, in such treatments the client traditionally enters a *transfer* phase. During the transfer phase procedures are introduced that are designed to generalise the newly learned speech skills to everyday speaking situations. The transfer phase can involve a whole new programmed instruction sequence involving speaking tasks that are arranged hierarchically, from easy to difficult.

## THE CLINICAL TRIAL EVIDENCE FOR SPEECH RESTRUCTURING TREATMENT

There have been more positive clinical trials of speech restructuring for persistent stuttering—more than 30— than for any other treatment. These trials are by many independent researchers in different countries, dating from 1973. A systematic review conducted in 2020<sup>23</sup> confirmed that this treatment for persistent stuttering has stronger evidence than any other.

### Effect size

As discussed during Lecture Five, effect size for a treatment can be assessed by comparing a treatment arm in a clinical trial with a control arm that receives no treatment. However, neither of the randomised trials of speech restructuring<sup>29,31</sup> involved a no-treatment control arm, so the effect size for the treatment cannot be estimated.

Another problem with knowing what the effect size might be for speech restructuring treatment is the high drop-out rates in clinical trials. In one trial<sup>32</sup> 20 of 32 dropped out, and in another trial<sup>28</sup> 13 of 30 dropped out. Equally troubling is that some trials with substantial participant numbers did not report whether there were any drop-outs: one trial with 36 participants,<sup>33</sup> one with 39 participants,<sup>34</sup> and another with 44 participants.<sup>35</sup> In fact, only one trial<sup>27</sup> with substantial numbers reported few drop-outs: two of 80 participants. So the usual bias where non-randomised trials overestimate effect size<sup>36</sup>

would be enhanced by these drop-out rates.<sup>†</sup> Only two clinical trials of speech restructuring treatment have used the technique of intention-to-treat analysis, mentioned during Lecture Five, to compensate for this problem.<sup>29,31</sup>

Considering all this, perhaps a reasonably conservative statement is that some of those who wish to attain clinically significant reduction of their stuttering will be able to succeed in doing so. The proportion of those who will succeed is unknown. The overall tenor of clinical trials—biased as they are—conveys that significant stuttering can be reduced to below 4 %SS or even below 1 %SS. There is some evidence that those stuttering severity reductions are not associated with changes to the behavioural complexity of stuttering.<sup>37</sup> As outlined during Lecture One, a stuttering moment can involve repeated movements, fixed postures, or superfluous behaviours. The report<sup>37</sup> indicated that, after speech restructuring treatment, there was no change in the overall behavioural complexity of stuttering moments. In other words, while the amount of stuttering reduced, the types of stuttering behaviours remained constant.

Again, with the reservation that the clinical trials concerned may be biased, it appears that clinically significant stuttering reductions may be obtained for periods of 1–2 years, which is the general follow-up period in clinical trials. The longest follow-up periods for clinical trials that showed sustained stuttering reductions were 9–12 years (N=12),<sup>38</sup> and 10 years (N=17).<sup>39</sup> Both results were for multi-week, intensive residential treatments.

As mentioned several times throughout these lectures, post-treatment relapse is a recurring problem with speech restructuring treatment.<sup>40,41</sup> Relapse rates were specifically reported in some of the clinical trials that have been published for adults. For example, one trial<sup>33</sup> reported 30–60% relapse at 12–18 months, depending on how relapse was defined. Another trial<sup>28</sup> reported 24% relapse in terms of more than 6.0 %SS at 12 months post-treatment.

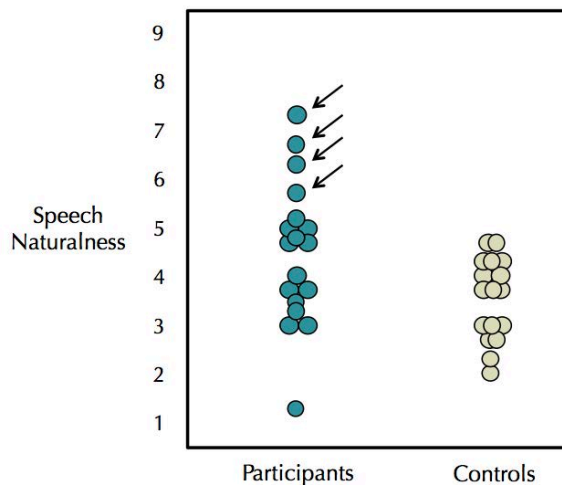
## Speech naturalness

### *How speech sounds*

As discussed during Lecture Four, gains from speech restructuring treatment are typically achieved at the cost of speech that does not sound perfectly natural. This problem has been known for decades.<sup>40</sup> There is no research evidence to support the contention, but it seems likely that unnatural sounding post-treatment speech contributes to the relapse problem.<sup>42,43</sup>

Communication effectiveness can be diminished if gains in fluency are achieved ... through the use of speaking techniques that are so burdensome and unnatural that the individual has difficulty using them on a consistent basis. (p. 290)<sup>43</sup>

Consequently, since the early 1990s, journal editors generally do not accept speech restructuring clinical trials for publication without speech naturalness assessment of some kind. The figure, \* from a clinical trial,<sup>44</sup> illustrates the issue. Listeners assigned speech naturalness (NAT) scores



<sup>†</sup> This reasoning assumes that drop-outs are connected to undesirable features of the treatment. However, it is possible that the reverse is true. Participants could drop-out from clinical trials because the treatment is so rapidly efficacious that they do not bother to continue their participation in the trial.

\* Adapted and reproduced with permission: O'Brian, S., et al. (2003). The Camperdown Program: Outcomes of a new prolonged-speech treatment model. *Journal of Speech, Language, and Hearing Research*, 46, 933–946. © 2003 American Speech-Language-Hearing Association.

to post-treatment speech samples of 18 participants who received speech restructuring treatment, and matched controls. As a group, the treated participants scored a little less than one NAT scale value higher than controls: means of 4.5 and 3.6 respectively. The figure shows that, except for four participants marked with arrows, the group who received treatment attained NAT scores around the range of controls.

### *How speech feels*

An important clinical issue with speech naturalness is that clinicians cannot assume that speech feels as natural to clients as it sounds. In fact, there is research to show that how natural speech sounds and how natural speech feels may be different things altogether.<sup>45,46</sup> The latter of those reports involved interviews of clients after speech restructuring treatment. Results showed one thing that drove clients to receive treatment was that they felt different because of their stuttering. However, the treatment by no means took away that feeling, but in fact worsened it: feeling different from their usual way of speaking. This is important information for clinicians. A treatment that controls stuttering but either sounds or feels unnatural may not be particularly useful to a client.

## **SPEECH RESTRUCTURING: THE CAMPERDOWN PROGRAM**

### **Background**

The Camperdown program is a publicly available speech restructuring treatment that is supported by clinical trials. For those reasons, it is outlined here.

Clinical resource materials are at the Camperdown Program website of the Australian Stuttering Research Centre.<sup>47</sup> The *Camperdown Program Treatment Guide* is a clinical reference for the treatment. Also at that website there are *Charts and Scales*, *Camperdown Program Training Examples*, and *Fluency Technique Samples*. If a clinician decides to use the program with an adolescent, few modifications are needed. Different versions of the audio training example, demonstrated by adolescents, are available on the Camperdown Program website.

As described earlier, speech restructuring typically involves teaching target speech behaviours: extended vowel production, light articulatory contacts, gradual onset of vocalisation, and continuous breath flow during speech. The Camperdown Program does not use any such speech targets. This is based on laboratory research questioning the value of such clinical methods.<sup>48,49</sup> The Camperdown Program does not involve any measures of stuttering severity.

The aim of the Camperdown Program is to give clients skills to control their stuttering to the extent that they find acceptable during everyday conversations. The following description of the seven treatment stages draws freely from the *Camperdown Program Treatment Guide*.

### **Stage 1: Camperdown Program Overview**

The clinician will have previously assessed the client's stuttering and related difficulties. Stage 1 is then an opportunity for the clinician to describe the program and its requirements. The clinician and client discuss individual goals and expectations for stuttering reduction.



## Stage 2: Learning the Fluency Technique

During Stage 2 consultations, clients learn to produce an individualised fluency technique using the *Camperdown Program Training Examples*. Clients listen to an audio recording of one or more of the training examples, and attempt to reproduce this fluency technique as closely as possible. At this Stage, the aim is to feel totally in control of stuttering while using the fluency technique. Clients are reassured here and throughout the early stages of treatment that this is a first step, and that natural-sounding speech is the purpose of treatment.

These are the steps for learning the fluency technique:

- (1) Introducing the fluency technique
- (2) Reading in unison with the training example
- (3) Reading the passage independently
- (4) Home practice with the training example
- (5) Using the fluency technique in different speaking tasks with the clinician
- (6) Independent practice using the fluency technique in different speaking tasks.

Clients progress to Stage 3 when they are able to use a very unnatural fluency technique, feeling completely in control of stuttering, while conversing with the clinician for 15–20 minutes.

## Stage 3: Refining the Fluency Technique

During Stage 3, clients gradually and systematically learn to shape unnatural-sounding stutter-free speech into more natural-sounding speech while still feeling in control of stuttering. The aim is to develop an individualised, reasonably natural-sounding fluency technique which controls stuttering while talking with the clinician. As yet, there is no expectation that this fluency technique will be used in everyday speaking.

These are the steps for refining the fluency technique:

- (1) Clients are shown the Fluency Technique Scale and introduced to the different amounts of technique.



### Amount of Technique



- (2) Learning and practising half technique. Clients listen to the clinician demonstrating half technique, and the audio examples of half technique. They gradually and systematically practise half technique in more difficult speaking tasks.
- (3) Learning and practising quarter technique. Clients listen to the clinician demonstrating quarter technique, and the audio examples of quarter technique, and then attempt to imitate these. Clients then practise quarter technique for other speaking tasks: reading passages, picture description, monologue, and conversation.

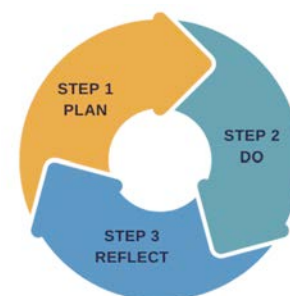
There is no expectation yet that quarter technique will be used in the real world. However, clients progress to Stage 4 when: speech sounds similar to the examples; quarter technique is used consistently during speaking tasks at home or in the clinic; and clients feel completely in control of stuttering when using quarter technique.

## Stage 4: Natural-Sounding Speech

During Stage 4, clients aim to sound as natural as possible while still controlling stuttering at a comfortable level. There is still no expectation that clients will use their fluency technique in everyday

speaking. Clients are introduced to minimal technique, speaking cycles, and the thinking process to improve their self-evaluation and problem-solving skills. The purpose of Stage 4 is to develop a sound, natural-sounding fluency technique as a foundation for stuttering reduction. Clients will take a while to find an individualised fluency technique they are happy with. There are no pre-determined targets; clients determine what they want to achieve in their everyday speaking. There are two components to Stage 4:

- (1) Minimal technique. The clinician introduces the concept of minimal technique by referring to the Fluency Technique Scale and demonstrating how minimal technique sounds. There are audio examples of minimal technique at the Camperdown Program website.
- (2) Speaking cycles. Clients are introduced to the three steps of a speaking cycle. Clients plan a speaking task, do the planned speaking task, then evaluate the result with a reflective process.



When clients understand the speaking cycles and the thinking process that underpins them, they can do speaking practice with a practice partner between clinic consultations. At the Camperdown Program website there is a speaking cycles form that can be used to assist clients to move through speaking cycles at home.

Clients progress to Stage 5 when they: practice speaking using a variety of fluency technique levels; typically use as much technique as planned; use fluency technique consistently; and when they are mostly in control of stuttering when using a comfortable amount of technique. This practice is still only with the clinician or a home practice partner.

### Stage 5: Preparing for Real-World Speaking

Stage 5 prepares clients for speaking in the real world by engaging in more complex conversations and attempting to use only minimal or quarter technique to control stuttering. Practice is still only done with the clinician or practice partner while sounding reasonably natural and still controlling stuttering. A practice partner is essential during Stage 5. The speaking cycles format outlined above is still used. The emphasis remains on self-evaluation and problem-solving skills. The clinician and client design challenging practice conversation tasks, keeping in mind the client's everyday interests and needs. Examples are: making a phone call to a familiar person; using technique during an extended conversation; moving from no technique, to minimal, to quarter technique; and having the practice partner talk quickly, talk over the client, or interrupting the client.

Clients progress to Stage 6 when they typically: use as much technique as planned; use the fluency technique consistently; are mostly in control of stuttering when using an acceptable amount of fluency technique; and would be comfortable talking like that in everyday situations.

### Stage 6: Natural-Sounding Speech

During Stage 6, clients learn to control stuttering in increasingly more challenging real-world conversations. The clinician prepares clients for the challenges that everyday conversations present; they are less predictable, there are more distractions, the content itself may be challenging, and clients may be anxious about stuttering or using their fluency technique. Clients are introduced to factors that may impact on speaking success, such as speech practice, cognitive demands, and anxiety. Clients learn ways to deal with these issues with an individualised hierarchy of speaking situations.

The number of Stage 6 consultations will vary according to stuttering severity, previous treatment, commitment to and completion of regular speech practice, regularity of weekly consultations, individual goals, and the presence of speech-related anxiety. Sessions become less frequent as the client gains confidence and success with everyday speaking.

A Stage 6 consultation usually consists of the following components, not necessarily in this order:

- (1) Confirmation of fluency technique
- (2) The speaking situation hierarchy

- (3) Client problem solving
- (4) Task deconstruction
- (5) Review and revision of practice routine
- (6) Discussion of the client's general stuttering control
- (7) Planning and summarising treatment changes.

### Stage 7: Maintaining Treatment Gains Long-Term

During Stage 7, consultations become less frequent according to client need. Clients learn to consolidate problem-solving skills to maintain, for the longer term, a balance between a comfortable amount of stuttering and the naturalness of fluency technique in everyday conversations. Clients also learn to deal with any increase in stuttering. Clients are reassured that when acquiring and using a new skill such as stuttering control, there are days when using that skill is easy and days when it is not. The three key points for keeping stuttering under control as needed are:

- (1) Having an appropriate speech technique practice routine
- (2) Being aware of factors that may influence the ability to control stuttering
- (3) Detecting and reacting quickly and appropriately to difficulties.

Discharge from treatment is negotiated between the clinician and the client when the client can demonstrate: attainment of personalised goals for stuttering treatment; sustained skills for monitoring speech and controlling stuttering; and the capacity to deal with fluctuations in stuttering control.

### Clinical trial evidence for the Camperdown Program

#### *Phase I and Phase II clinical trials*

Using the Lecture Five definition of a clinical trial, there have been several supportive Phase I and Phase II trials of the Camperdown Program speech restructuring treatment model. Three trials involved a standard clinic treatment format,<sup>50,44,51</sup> (the latter with adolescents) and one reported results at a university student clinic.<sup>52</sup> Another was a low-tech telephone telehealth trial with adults,<sup>53</sup> and another was a video telehealth trial with three adolescents.<sup>54</sup>

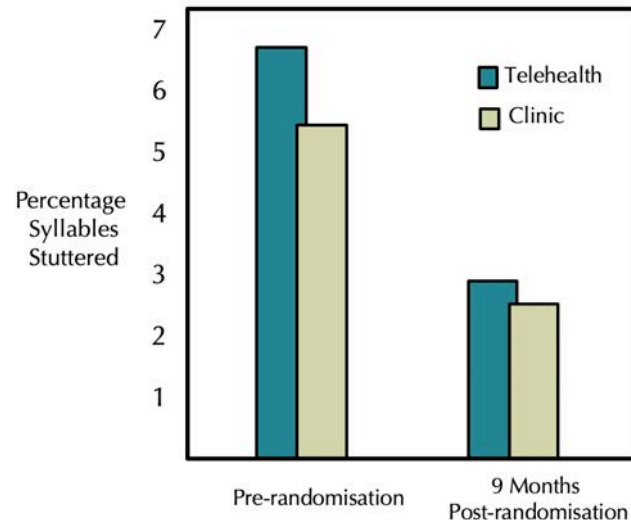
One report involved an experimental version of a standalone internet presentation of the treatment that did not require a clinician.<sup>55</sup> The results of this standalone internet Phase I trial with two adults was encouraging. A subsequent trial of the standalone internet version recruited 20 adults.<sup>56</sup> As with all clinician-free internet treatment programs, compliance was an issue. Five participants completed the treatment and five completed more than half of it. Four of the five who completed the treatment reduced their stuttering severity by more than half, and two of those who completed more than half the treatment reduced their stuttering severity by an equivalent amount. Results were confirmed by participant reports of stuttering severity. These results suggest that standalone internet Camperdown Program treatment may be a useful component of the stepped-care approach to stuttering described during Lecture Six.

#### *A Phase III clinical trial of telehealth*

As mentioned during Lecture Six, there are some compelling advantages for telehealth treatment services with early childhood stuttering. In the case of adults, there is the advantage for the many young adults who wish to reduce their stuttering for employment reasons and are reluctant to take time off work. In such situations, telehealth treatment can minimise work disruption by reducing travel time to the clinic. Additionally, consultations within the client home maximise cultural and community support during treatment.<sup>57</sup> There is evidence that video telehealth assessment of adults is viable<sup>58</sup> (see Lecture Four).

A randomised Phase III trial<sup>29</sup> compared the standard clinic Camperdown Program presentation with an experimental, low-tech telehealth version presented by telephone. For the telehealth adaptation, “home practice replaced the face-to-face programme group intensive day” (p. 110).<sup>29</sup> The trial used a non-inferiority design, which establishes whether an experimental treatment variation is not inferior to the original. Twenty adult participants were recruited to each arm of the trial.

Results are presented in the figure.\* The telehealth group had more severe stuttering than the group that received the standard, in-clinic treatment. This can occur with small participant numbers, even though participants are randomised to each group. Three participants dropped out (8%) and their data were analysed by intention-to-treat with last observation carried forward (see Lecture Five). After treatment there was no difference in outcomes between the two groups, and in fact it is arguable that the telehealth group did better, considering that their stuttering was more severe pre-randomisation.



The telehealth group required a mean of 10 hours 17 minutes for treatment, and the standard group required 12 hours 54 minutes. When these values were statistically adjusted for differences of pre-treatment variables such as stuttering severity, prior treatment and family history, the telehealth group used 221 minutes less contact time—3.7 hours—than the standard group. That result was clinically significant and statistically significant.

Speech naturalness assessment used a control group as a reference. There was a statistically significant result where both treatment groups had a mean speech naturalness score one scale value less natural than control speakers. In other words, to some extent, control of stuttering was attained at a cost of speech that sounded unnatural to some extent. These results were similar to those for a Phase I trial of the Camperdown Program.<sup>44</sup>

#### *No replications*

This body of clinical trial evidence might be interpreted as a sound data base showing the efficacy of the Camperdown Program model. However, there has been no independent replication of any of these results, and all the trials cited previously were from the same research group, albeit from researchers located in three different cities. As such, the results require cautious interpretation.

## **SPEECH RESTRUCTURING: THE COMPREHENSIVE STUTTERING PROGRAM**

### **Overview**

This evidence-based treatment model is outlined as a contrast to the Camperdown Program. It is a 3-week residential treatment that incorporates speech restructuring targets, programmed instruction, and a transfer phase. The Comprehensive Stuttering Program was developed during the 1980s<sup>59</sup> and is provided as a 90-hour intensive treatment service at the Institute for Stuttering Treatment and Research<sup>60,61</sup> in Edmonton, Canada. It appears that a freely available treatment manual is not available to the public. A 2007 description of the treatment is available.<sup>62</sup> However, it is not clear what

\* Adapted and reproduced with permission: Carey, B., et al. (2010). Randomized controlled non-inferiority trial of a telehealth treatment for chronic stuttering: The Camperdown Program. *International Journal of Language and Communication Disorders*, 45, 108–120. © 2010 Taylor & Francis.

developments have occurred since then, although the website states that “the program is continually evaluated and improved.”<sup>60</sup> Because it is supported by clinical trials and is currently provided as a treatment service, it is outlined here.

Although the 3-week residential intensive treatment model can be adapted as needed, “it is the preferred format for the majority of clients” (p. 214).<sup>62</sup> This intensive format involves 90 hours of therapy with 6 hours per day. The Comprehensive Stuttering Program has three standard, formal phases: acquisition (instatement), transfer, and maintenance. The transfer phase involves a series of beyond-clinic speaking tasks, including speaking to strangers, telephoning businesses, shopping assignments, and group presentations.

The programmed instruction sequence begins at 40 SPM and with an eventual target of 190 SPM +/- 40 SPM. The speech pattern is taught using the following speech targets: “prolongation,” “easy breathing,” “gentle starts,” “smooth blending,” and “light touches” (p. 217).<sup>62</sup>

These speech targets are taught with constant attention to attaining natural sounding speech, and clients learn to use a 10-point speech naturalness scale, which is used during the treatment process. The treatment incorporates Van Riper’s “stuttering modification” techniques (“stutter more fluently”) that were described earlier.

The Comprehensive Stuttering Program involves a substantive nonbehavioural component with cognitive behaviour therapy for each client (a psychological intervention: see Lecture Twelve). The Comprehensive Stuttering Program does not include any standard clinical psychology measures, and it appears that speech-language pathology staff, rather than clinical psychologists, administer the cognitive behaviour therapy component.

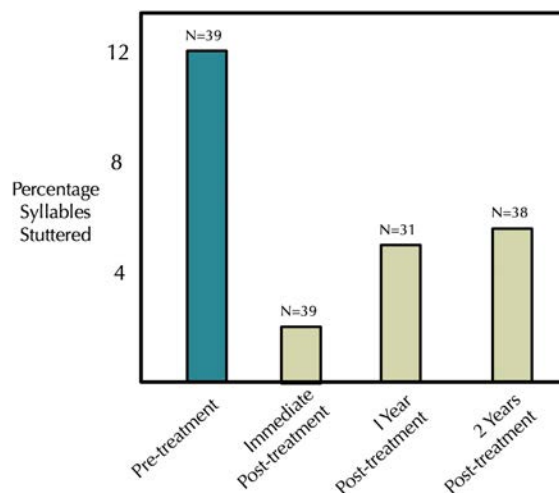
### Clinical trial evidence for the Comprehensive Stuttering Program

As with the Camperdown Program, there has yet to be an independent replication of clinical trial results for this treatment. Phase I and Phase II clinical trials were published in the 1990s,<sup>28,63</sup> the former involving 42 participants, 17 of whom were adults and 25 were adolescent. Subsequently, a Phase II trial was reported<sup>34</sup> involving 39 participants, 14 of whom were Canadian and 25 were Dutch. Although the trial purports to be for adults, the age range of the Dutch group was 17–53 years, and the range for the Canadian group was 15–42 years. So, a small, unknown number of participants were adolescents.

Speech measures were made pre-treatment, immediately post-treatment, and at 1 and 2 years post-treatment. At 2 years post-treatment there was only one drop-out from the study. Data were not available for eight participants at 1 year post-treatment. Results are presented in the figure, with the Canadian and Dutch participants pooled.

Speech naturalness data were presented in this clinical trial but are difficult to interpret. The Dutch participants’ speech naturalness was measured with a procedure not common in stuttering clinical research: a 7-point bipolar naturalness scale.

The standard speech naturalness (NAT) measure was used for the Canadian participants, however participant speech samples were presented to listeners along with “140 other speech samples” (p. 238).<sup>34</sup> No indication was given of the nature of those speech samples and how they may have influenced NAT scores for the trial participants. With this reservation, the mean NAT score at 2 years post-treatment was 2.9, which certainly suggests natural sounding speech. However, mean speech rate





data presented in the report (Table 1, p. 241)<sup>34</sup> are 135 SPM at 2 years post-treatment for the Dutch participants and 153 SPM for the Canadians. The target SPM range for the Comprehensive Stuttering Program treatment process is 190 SPM +/- 40 SPM, with “most clients ... speaking at a rate of 150 to 190 SPM, which is on the lower end of the normal range” (p. 219). This might be interpreted to suggest that Table 1 (p. 241) of the report shows unusually slow speech and, hence, unnatural sounding speech.

In another report with one of the longest follow-up periods on record,<sup>39</sup> the same research group reported that at 10 years post-treatment 17 participants, when telephoned unexpectedly, showed that they had retained similar treatment effects to the participants who were studied at 2 years post-treatment.<sup>34</sup> No speech naturalness data were presented to bolster confidence in this result. However, self-report data indicated that

at 10 years follow-up the majority of participants who responded reported that (1) they were generally satisfied with their current speech, (2) they had the ability to use techniques to control speech most of the time or more often, (3) their confidence in their ability to speak improved and (4) they had to pay attention to speech most of the time or almost always to be fluent. (p. 120)<sup>39</sup>

## **SPEECH RESTRUCTURING: VIDEO SELF-MODELLING AS A SUPPLEMENT**

### **The procedure**

Self-modelling is positive behaviour change based on people observing themselves being free of a problem behaviour. It has been shown useful for managing various behaviours,<sup>64</sup> and, in theory, can be used as an additive to improve the effects of any stuttering treatment. Clinicians can make a video recording of clients speaking without stuttering, using the speech restructuring technique. Then, simply, clients are instructed to watch the videos regularly.

### **Basic research**

The mechanisms by which self-modelling might work are unclear, but there has been a suggestion that it relates to self-efficacy and self-belief.<sup>65,66</sup> With stuttering, two single-subject laboratory experiments<sup>67,68</sup> showed promise for children and adolescents, with encouraging results continuing at 2–4 years follow-up.<sup>69</sup> Subsequently, a laboratory study<sup>70</sup> was conducted with three adults using an experimental single-subject design. The researchers made self-modelling videos of the participants by having them repeat any stuttered utterances until they were stutter-free, and then editing the stuttering moments from the videos. During the experimental condition participants were instructed to speak the way they did on the videos. One participant showed clinically significant stuttering reductions under that condition.

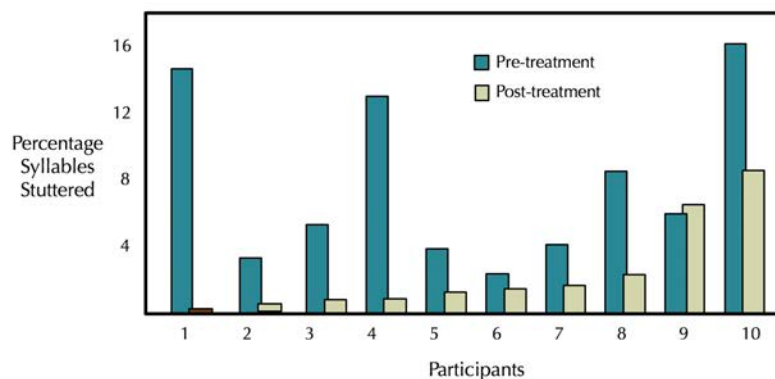
### **A data-based case study of video self-modelling: Relapse management**

#### *Design*

A case study report<sup>71</sup> explored the potential value of video self-modelling with the common relapse problem after speech restructuring stuttering treatment. The study recruited 12 adults who had received speech restructuring treatment but had relapsed. The researchers gave them a 1-hour clinical session during which the participants re-established speech that was stutter-free and as natural sounding as possible. For each of the participants, three 5-minute videos were constructed that contained no stuttering. The mean NAT score for them was 3.8. The participants were asked to watch the videos once per day for 1 month. Twelve participants were recruited and two (17%) dropped out.

### Results

Results are presented in the figure,\* which shows pre-treatment and post-treatment stuttering severities immediately after the 1 month of watching the videos. Participants 1–8 restored their stuttering severities levels to those that are associated with successful speech restructuring treatment outcomes. Participant 10 reduced severe stuttering by around half and one participant did not respond at all. The mean post-treatment NAT score was 3.9, which was comparable to the result for another speech restructuring treatment by the same research group,<sup>44</sup> suggesting that the regaining of stuttering control required some compromise of speech naturalness.



### Clinical applications

This finding suggests that with just an hour of clinical time spent, clinicians can successfully manage clients who have relapsed after speech restructuring treatment. Additionally, the technique could be used with the intention to prevent the occurrence of relapse. This is potentially far more efficient than the common “booster” or “refresher” sessions that are reported in descriptions of speech restructuring treatments,<sup>62,72</sup> where clients return to the clinic to receive a substantial portion of their treatment to restore their speech benefits, or to provide resistance to relapse occurring.

## A Phase III trial of video self-modelling: Speech restructuring supplement

### Design

The Phase I trial results of video self-modelling led those researchers to explore whether building video self-modelling into a speech restructuring treatment process might improve outcomes. So, 89 participants, 64 adults and 25 adolescents, were recruited into a Phase III trial.<sup>31</sup> One arm involved standard speech restructuring, and the experimental arm involved standard speech restructuring plus video self-modelling. The participants were treated at different clinics around Australia, using different speech restructuring treatment models.

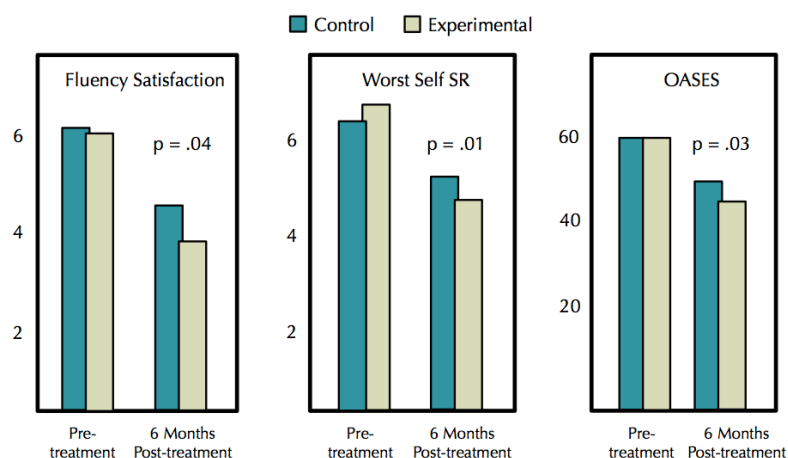
Towards the end of the treatment, the researchers made 4.5-minute self-modelling video recordings of all clients. Clients in the experimental group were given their self-modelling video and instructed to watch them for 5 minutes each day for a month, with the following instruction: “Try to talk without stuttering using your speech technique as you see yourself doing on the video” (pp. 890–891).<sup>31</sup> Five participants (6%) dropped out, and their results were analysed with intention-to-treat analysis by last observation carried forward (see Lecture Five).

The primary outcome was %SS measured from unscheduled telephone calls to participants from strangers. There were several secondary outcomes: Subjective Units of Distress<sup>73</sup> scores for anxiety after the telephone calls, self-rated SR scores for eight nominated speaking situations, avoidance of those eight speaking situations, satisfaction with fluency, and impact of stuttering measured with the OASES (see Lecture Four).

\* Adapted and reproduced with permission: Cream, A., et al. (2009). Self-modelling as a relapse intervention following speech-restructuring treatment for stuttering. *International Journal of Language and Communication Disorders*, 44, 587–599. © 2009 Taylor & Francis.

## Results

Three significant results were found: fluency satisfaction, improvement in the speaking situation self-rated most severely, and the OASES. The OASES scores for the experimental group dropped from moderate impact at pre-treatment to mild-moderate impact at post-treatment. Results are presented in the figure below. The paper does not present data separately for adult and adolescent participants, although the authors state that there were no differences in outcomes between the two age groups. So, the figure, although it represents adults and adolescent results combined, is likely to be reasonably representative of the adult group results.



There can be some flexibility with incorporating the results of the two trials of video self-modelling into clinical practice. For example, a clinician may require clients to watch self-modelling videos daily for a month after treatment and then systematically withdraw how often they watch them until they can manage to sustain their treatment benefits without any watching. Then, for impending relapse, a client could return to daily watching. Only when a return to daily watching fails to prevent signs of impending relapse would a client need to come to the clinic for further consultation.

## A clinical experiment

Participants in this experiment<sup>74</sup> were three adults who had received an intensive, residential speech restructuring treatment.<sup>62,75</sup> They were studied with a multiple baseline across participants experiment, which is one of many experimental designs used with individuals. Prior to the experiment, their stuttering severities were 6.4 %SS, 7.7 %SS and 16.2 %SS. Each of them was given two 4-minute self-modelling videos, which they were instructed to watch at least twice per week for 5 weeks. Stuttering severity during the 5-week experimental period was measured each week with three recordings, one of which was independent of the clinic. The researchers reported that two of the participants reduced their stuttering severity. One participant reduced %SS scores by around one fifth, and another by around a third. However, the data are difficult to interpret because the reported %SS scores were an amalgam of within- and beyond-clinic measures. All three participants reported that the self-modelling procedure benefited them: “the most robust improvements in the self-report data were reduced avoidance behaviors followed by reduced expectancy to stutter” (p. 39).<sup>74</sup>

## SUMMARY AND CONCLUDING COMMENTS

### Limitations of speech restructuring

As noted during Lecture Five, many clients require behavioural control of stuttering. Although speech restructuring is a long-standing treatment which is acceptable to many as one way to do so, it has limitations and is far from a perfect clinical solution. Speech restructuring may control stuttering at the cost of speech that sounds unnatural to the listener or speech that feels unnatural to the speaker, or both. The complexities of coordinating laryngeal, respiratory, and articulatory processes during speech are of little conscious concern to most. But speech restructuring treatment requires attention to those speech processes to control stuttering. That requires considerable effort and practice. And during Lecture Ten the issue is raised that speech restructuring treatment may not be ideal for those who stutter and who are socially anxious.

### Adults

According to clinical trials, speech restructuring is the most efficacious speech treatment for persistent stuttering during adulthood. For some adults, despite its many limitations, the treatment is clearly capable of controlling stuttering to a clinically significant extent and for a clinically significant period. Yet currently, without randomised controlled evidence, nothing is known about effect sizes that might be expected. There is convincing evidence that video self-modelling is a useful adjunct to speech restructuring treatment.

### Adolescents

For adolescents, speech restructuring has the strongest supportive clinical trial evidence of all available treatments for that age group, with independent replications. There is evidence that video self-modelling can be useful with this age group. Arguably, considering the potential benefits against the little effort required to implement a video self-modelling procedure, it should be a routine adjunct during and after a speech restructuring treatment process with adolescents. It is also necessary to note that the major drawback of this style of treatment—speech that sounds and feels somewhat unnatural—may be a particular clinical issue with adolescents.

### School-age children

For school-age children, there is little evidence for speech restructuring. One clinical trial of an intensive treatment for 9–14-year olds has been reported,<sup>76</sup> but it was not overviewed in this lecture because a treatment manual is not publicly available, and it does not appear to be used any longer. The same reservation applies to a data-based case study<sup>77</sup> of an intensive speech restructuring treatment with 6–8-year-olds. It is of interest that a systematic review<sup>78</sup> identified an overlooked data set that included a variant of speech restructuring for 10 children in the age range of 5–12 years.<sup>79</sup> Those data included the Perceptions of Stuttering Inventory, which is a scale of anxiety that is related to stuttering. The review suggested that the speech restructuring variant may have improved anxiety for this age group without any specific procedures for anxiety management.

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## LECTURE NINE: OTHER EVIDENCE-BASED SPEECH TREATMENTS FOR PERSISTENT STUTTERING

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### THE LIDCOMBE PROGRAM

#### Case reports

Although the Lidcombe Program was developed for children younger than 6 years, there are data supporting its use with older children. A follow-up of eleven 6–10-year-old children who were treated with the Lidcombe Program showed positive results.<sup>1</sup> The children were telephoned three times during one week at a mean of 70 weeks post entry to Stage 2, with a range of 9–187 weeks. At that follow-up their mean %SS score was 1.9. The results also are consistent with a positive data-based case report of the Lidcombe Program with a bilingual Iranian school-age child.<sup>2</sup>

#### Two Phase II trials

##### *An early in-clinic trial*

An early Phase II trial of the Lidcombe Program<sup>3</sup> with school-age children recruited 15 Australian families and treated them with a standard in-clinic weekly-visit format. Four of the families dropped out, and the remaining children had a mean age of 8.3 years with a range of 6.10 years to 12.4 years. The trial showed a stuttering reduction of 89% from a mean pre-treatment stuttering severity of 5.3 %SS. To reach Stage 2, a mean of 15 hours of treatment was required, with a median of 12 hours. The report contained no speech naturalness assessments, perhaps with the assumption that speech naturalness would not be an issue with a verbal response contingent stimulation treatment.

##### *A later video telehealth trial<sup>†</sup>*

A more recent Phase II trial of the Lidcombe Program with school-age children<sup>4</sup> recruited 43 6–12-year-olds from Singapore, Hong Kong, New Zealand, and Australia. Six of the families dropped out shortly after beginning the treatment. Treatment was delivered with video telehealth, following the Lidcombe Program Treatment guide, but with modifications designed to suit and engage school-age children.

The primary outcome was stuttering severity, based on beyond-clinic recordings of children speaking with parents, non-family members, or with a friend on the phone. An independent clinician measured stuttering severity from these recordings using a 10-point scale. Secondary outcomes were stuttering impact measured with the OASES-S (see Lecture Four), the Communication Attitude Test (see Lecture Twelve), and the Spence Children's Anxiety Scale (see Lecture Twelve).

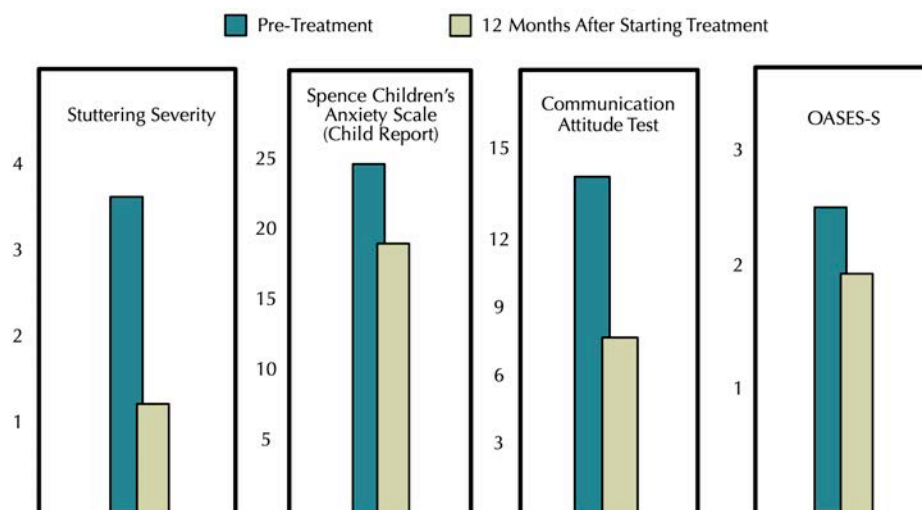
For the 37 children who completed their treatment, there was a mean stuttering severity reduction of 67%, with most children reducing stuttering severity by one scale value from a pre-treatment mean of 3.6. Of the 37 children, 32% (n=12) attained Stage 2 of the Lidcombe Program, with no stuttering or nearly no stuttering. More than half the children The mean scores for the 37 children for stuttering impact, communication, and anxiety, all showed clinically significant reductions. Results are presented in the figure below. The authors concluded that, although nonrandomised evidence is likely to overestimate effect sizes, parents can be informed that, for school-age children, “the Lidcombe



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<sup>†</sup> Techniques for adapting the treatment for 6–12-year-olds are overviewed later in this lecture under the heading “Clinical notes about persistent stuttering: School-age children.”

Program provides a chance of halting further stuttering development" (p. 8).



## THE OAKVILLE PROGRAM

### Resource materials

These are located at the website of the Australian Stuttering Research Centre.<sup>5</sup> The *Oakville Program Treatment Guide* is a clinical reference for the treatment. There is a clinical severity rating (SR) chart for parents and clinicians: *Child Stuttering Severity Chart eForm*. To assist clinicians to deliver this treatment, the website contains a *Syllable-Timed Speech Training Model* and four *Video Demonstrations of Syllable-Timed Speech*. There is also the *Stuttering Treatment Activity Guide for School-age Children*.

### A Phase II trial

The researchers who published a Phase I trial of syllable-timed speech for children<sup>6</sup> sought to improve the modest result in that trial of mean stuttering reduction of around 50%. They proposed that a way to do that might be to add parent verbal contingencies to the treatment.<sup>7</sup> The hybrid treatment is essentially the same as the syllable-timed speech treatment,<sup>6</sup> except that during Stage 1, after syllable-timed speech practice is introduced at home, parent verbal contingencies for stuttering and stutter-free speech are introduced.

Twenty-two children, aged 6–11 years, were recruited to the trial, 16 of whom were boys and six were girls. Fourteen of the children had previous treatment, and nine had comorbid speech and language conditions. Outcomes were measured pre-treatment and 6 and 12 months after completion of Stage 1.

### Results

#### *Stuttering severity*

Three children withdrew from the trial before completing Stage 1, leaving 19. Results are presented in the figure.\* The mean reduction of %SS scores at 12 months post Stage 1 completion was 77%. The mean %SS score at 12 months post Stage 1 was 1.9, with a range of 0.2–5.6 (SD=1.6). At each assessment the children reported their typical stuttering severity in each of the eight situations used in the Phase I trial.<sup>6</sup> The group mean pre-treatment typical stuttering severity (not presented in the figure)

\* Adapted and reproduced with permission: Andrews, C., et al. (2016). Phase II trial development of a syllable-timed speech treatment for school-age children who stutter. *Journal of Fluency Disorders*, 48, 44–55. © 2009 Elsevier.

was 5.4 and 1.9 (SD=1.2) at 12 months post Stage 1.

### *Impact of stuttering*

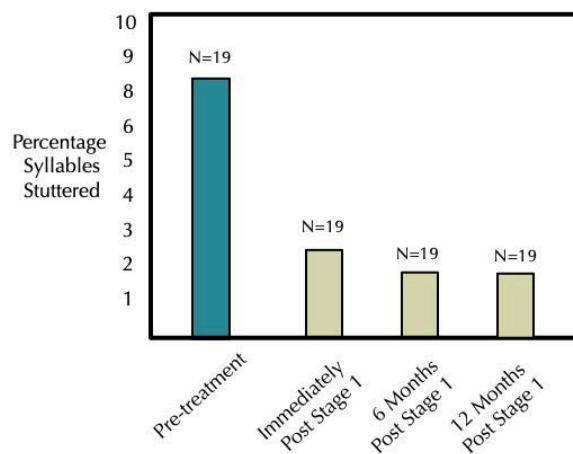
At each assessment the children completed the Assessment of the Child's Experience of Stuttering, which is an earlier version of the OASES-S (see Lecture Four). The mean pre-treatment score was 50.6 and 33.9 at 12 months post Stage 1. This change represented an improvement from moderate to mild-moderate impact.

### *Avoidance*

As discussed in Lectures One and Ten, situation avoidance is common with stuttering, and is associated with social anxiety, as considered during Lectures Ten and Eleven. Situation avoidance was measured at each assessment with a three-point scale of how often the children avoided eight speaking situations: "never," "sometimes," or "usually." The scores were converted to 0–2 values, with a maximum score of 16 for the eight speaking situations. A group reduction was found from 5.7 to 2.1. A systematic review of psychosocial effects of treatment for children ages 6–12 years<sup>8</sup> indicated that this result is promising; it suggests that a behavioural treatment might improve anxiety for this age group without any specific procedures for anxiety management.

### *Post-treatment speech*

To determine if listeners could detect any signs of speech rhythmicity post-treatment, 10 seconds of stutter-free speech pre-treatment and 10 seconds post-treatment was selected for each child. A group of listeners was asked to score each sample with a five-point scale where 0 = *not at all rhythmic speech*, and 4 = *extremely rhythmic speech*. Results showed no sign that the children were speaking with any detectable rhythm post-treatment.



## DELPHIN SPEECH TREATMENT

### Method

The methods of this German treatment for children and adolescents are outlined in the one available English report about it.<sup>9</sup> Although it is not fully clear, the treatment seems to incorporate elements of speech restructuring:

The first goal is the acquisition of costo-abdominal breathing ... Then the patient is taught in one-on-one-sessions what Schültz calls the "deblocking impulse," a kind of sigh, a letting go. The deblocking impulse is formed without pressure. It leads to a relaxation of the region around the larynx and impedes a cramping of the false and true vocal folds. This deblocking impulse should be used in every syllable with the main accent ... Approximately, 5 days later, the second step follows. Now the patient learns what Schültz calls "nasal swinging accent": a gentle voice onset with nasal character in words beginning with a vowel. The nasal character facilitates a soft voice onset. (p. 159)<sup>9</sup>

There is no treatment protocol available to provide more precise details about this treatment, but it incorporates progressive muscular relaxation, and the unusual inclusion of

... European drum sessions to foster sense of community and to improve concentration and coordination skills. In contrast to African drum playing, where the right hand is mainly used, here the player alternates from one hand to

the other after each beat; both halves of the brain are activated and are said to be better connected. During the drum sessions, the speech technique is used, and for some patients the movement of the hands facilitates the speech technique. (p. 160)

### A Phase II trial

Fifty-six participants were recruited with a median age 13.0 years, 42 of whom were boys. Inexplicably, a small but unknown number of adults were included, and the age range of participants was 8–36 years. Participants were treated in groups of seven to 10. The treatment duration details are not clear apart from the fact that it was resource intensive: “during the intensive therapy, a patient attends 140 sessions (mostly group sessions),” and “during the 2 years after the intensive therapy, the patient has maximally 2 weeks and four single days for the stabilisation phase, usually 50–80 logopedic sessions” (p. 160).<sup>9</sup> Various speech measures were collected at pre-treatment and at various times up to 12 months post-treatment, and one of those measures involved a telephone call to participants outside the clinic. The Strengths and Difficulties Questionnaire<sup>10</sup> was used to assess emotional and behavioural problems pre-treatment and post-treatment.

### Results

#### *Stuttering severity*

It is difficult to interpret the results of %SS scores for pre-treatment and post-treatment telephone conversations, because they were based on short samples of 250 syllables. The pre-treatment mean was 12.7 %SS and the post-treatment mean was 4.3 %SS.

#### *Speech naturalness*

A five-point scale was used to evaluate whether the parameters “prosody,” “breathing,” and “tonus during phonation” were “normal” or “markedly deviant.” Although improvements were reported, it is not possible to determine the extent to which participants sounded natural at post-treatment.

#### *Strengths and Difficulties Questionnaire*

Improvements were reported from pre-treatment to post-treatment. However, it is not possible to attribute those improvements to the speech treatment component or to non-speech treatment components (progressive muscular relaxation and group based activities such as “drum sessions”).

## A DATA-BASED REPORT OF AN INTENSIVE UNIVERSITY CLINIC

An intensive treatment at Bowling Green State University in the United States is conducted by speech-language pathology students under supervision.<sup>11</sup> The clinic involves 60 hours of in-clinic treatment, with “a minimum clinician-to-client ratio of 1:1” (p. 257) followed by twice-weekly, 1-hour telehealth follow-up sessions for 10 months. The treatment combines elements of cognitive behaviour therapy (a psychological intervention: see Lecture Eleven), speech restructuring, and elements of Van Riper’s “stuttering modification” techniques (see Lecture Eight):

- (1) education about speech production and stuttering;
- (2) awareness of stuttering behaviors, thoughts, and feelings;
- (3) desensitization to the moment of stuttering;
- (4) desensitization to listener reactions;
- (5) increased fluency via fluency-shaping strategies; and
- (6) reducing stuttering severity via stuttering modification techniques. (p. 257)<sup>11</sup>

A retrospective, data-based case report of 17 adolescents and adult clients (mean age 22 years, range 11–42 years) who received this treatment indicated improvements at 10 months follow-up. Mean %SS scores changed from 7.3 pre-treatment to 4.5 at follow up, and OASES scores (see Lecture Four) changed from moderate to mild-moderate impact. It is difficult to interpret these results, for several reasons. The treatment was provided at a student clinic, and the %SS scores were obtained from within-clinic conversations with treating clinicians.

## KINDER DÜRFEN STOTTERN (KIDS)<sup>†</sup>

### The treatment

This stuttering modification treatment for school-age children was developed in Germany, based on Van Riper's speech modification approach (see Lecture Eight).<sup>12</sup> The treatment manual<sup>13</sup> is in German, and details of the treatment are available in English Supplemental Material to a clinical trial report of the treatment.<sup>14</sup> There are five phases to KIDS: Information and Contract, Desensitization, Identification, Modification, and Generalization.

During the *Information and Contract* phase, the parents and child are informed about stuttering, its consequences, and the treatment plan. The child is educated about stuttering, and a therapy contract is established between the child, the parents, and the clinician. The *Desensitization* phase focuses on anxiety, targeting reduction of avoidance behaviour, talking openly about stuttering, and responding appropriately to negative listener reactions. This phase involves pseudostuttering, also known as voluntary stuttering (see Lecture Eight), which is designed to reduce anxiety about stuttering. Children learn to do this initially in the clinic using a sequence beginning with sentences and leading up to the procedure being used in everyday situations. The *Identification* phase focuses on analysing the child's stuttering behaviours and reactions to them. The child is trained to monitor for stuttering moments when speaking and to reduce avoidance behaviours. The *Modification* phase involves children using Van Riper techniques to deal with stuttering moments. They use the "pullout" technique in everyday situations to deal with stuttering moments. They also use "prolongation," which appears to be the Van Riper "preparatory set" technique. During the *Generalization* phase children learn to transfer skills to everyday life, and clinic visits become less frequent.

KIDS is presented with one or two 45-minute clinic sessions per week. Treatment ends when:

- (a) the child stutters mildly (symptoms last less than half a second and are without associated struggle behaviors) or not at all; (b) the child has a positive self-efficacy in coping with stuttering symptoms, difficult speech situations, and negative listener reactions related to stuttering; and (c) adequate reactions in the environment toward stuttering prevail (p. 3).<sup>14</sup>

### A Phase II trial

Participants in this trial<sup>14</sup> were children 7–12 years old who were diagnosed as stuttering by physicians and referred for treatment. They were randomised to an immediate treatment group and a waiting list control group. After 3 months, the children in the control group began treatment. The authors argued that 3 months of withholding treatment from this age group was the maximum that could be ethically justified. Both groups received 12 months of treatment. Thirty-seven children were randomised to the immediate treatment group, and 36 were randomised to the control group. Thirty-three children in the immediate treatment group completed 12 months of treatment, and 26 in the control group completed 12 months of treatment. Twenty-six clinicians conducted the treatments.

The primary outcome was OASES-S total scores. Secondary outcomes included parent severity ratings and parent "satisfaction with the child's ability to communicate" (p. 5) using a 10-point scale. Other secondary outcomes included total SSI-4 scores (see Lecture Four) from a 15-minute within-clinic recording, and %SS scores from beyond-clinic recordings of the children with a parent and an unrelated adult.

### Results

At 3 months, there was a significant difference in the primary outcome of OASES-S total scores. At that time, the children in the immediate treatment group had received a mean of 11 treatment sessions. OASES-S scores changed by 0.38 in the immediate treatment group and 0.12 in the control group. No changes in secondary outcomes were reported at that time. The latter result was not surprising,

<sup>†</sup> "Kinder Dürfen Stottern" translates to "children are allowed to stutter."

considering that only 6% of children had completed their treatments after 3 months.

After the randomisation part of the trial, all children's outcomes were reported for the longer term as one group, with a mean of 36 treatment sessions over 39 weeks. After 12 months of treatment, 62% of the children had completed their treatments. Significant effects were found for most secondary measures. Mean OASES total scores changed from 2.4 to 1.9, representing a change from moderate to mild-moderate impact. Mean parent severity ratings changed from 4.5 to 3.5, and mean parent satisfaction with communication scores changed from 7.4 to 8.4.

## **A MACHINE-AIDED TREATMENT: NONINVASIVE BRAIN STIMULATION**

Noninvasive brain stimulation is a term describing clinical methods "to modulate the excitability of the brain via transcranial stimulation" (p. 173).<sup>15</sup> The two methods are transcranial magnetic stimulation and transcranial direct current stimulation.

### **Transcranial direct current stimulation**

Transcranial direct current stimulation "involves application of a weak electrical current across the head via electrodes placed on the scalp, modulating the resting membrane potential of neurons in the underlying cortex" (p. 2).<sup>16</sup> The technique has been used, with mixed results, for a range of conditions, particularly psychiatric illnesses, and for cognitive enhancement with healthy participants. This treatment development was spurred by applications of the technique to rehabilitation of limb and speech motor function with stroke patients. There have been feasibility studies with stuttering,<sup>17,18,19</sup> and a case study report, with one participant, of transcranial direct current stimulation as an adjunct to speech treatment.<sup>20</sup> There is a detailed review of eight studies of the technique applied to stuttering reduction in adults.<sup>21</sup>

### **Case report: An adjunct to chorus reading and syllable-timed speech**

A randomised clinical experiment was reported<sup>16</sup> with the technique as an adjunct. Thirty participants were randomised to receive either five consecutive days of 20-minute speech treatments with transcranial direct current stimulation, or five consecutive days without that adjunct. The speech treatment was—inexplicably—"choral speech, and metronome-timed speech" (p. 4). Participants who did not receive the transcranial direct current stimulation received a sham treatment involving a dosage that was presumed to be ineffective. The 20-minute speech treatment sessions comprised a sequence designed as a hierarchy of increasing difficulty: chorus reading with a clinician, chorus reading with recorded speech, syllable-timed speech with monologue, and syllable-timed speech in conversation.

The primary outcome was change in "percentage of disfluent syllables" from pre-treatment to 1 week post-treatment, and from pre-treatment to 6 weeks post-treatment. At each assessment, the measure was generated from clinic speech samples of reading and conversation. The first 2 minutes of each sample was analysed. Thirty participants received treatment after randomisation. The control group showed no change. Overall, for the experimental group, there were changes from pre-treatment percentage disfluent syllables scores: 27% reduction at 1 week post-treatment and 22% reduction at 6 weeks post-treatment. However, at 6 weeks post-treatment, although gains remained for the reading task, the percentage of disfluent syllables score for conversation had returned to pre-treatment levels. Perhaps controversially, considering that their data suggested otherwise, the authors concluded that "transcranial direct current stimulation combined with behavioural fluency intervention can improve fluency in adults who stutter" (p. 1161).<sup>16</sup>

A smaller study involved 10 participants in a crossover clinical trial design with two conditions: active or sham transcranial magnetic stimulation, each combined with syllable-timed speech training.<sup>22</sup> Training involved 30 minutes on five consecutive days in one week on one condition, and a crossover to the other condition after 2 weeks. A week later there were speech improvements in both groups, but no significant effects for transcranial magnetic stimulation.

### Case report: An adjunct to delayed auditory feedback

A report involved transcranial direct current stimulation as an adjunct to delayed auditory feedback.<sup>23</sup> The reason for the choice of delayed auditory feedback, rather than a treatment known to be more effective, was not clear: the report states, correctly, “that the effectiveness of DAF for the reduction of stuttering is limited in everyday speaking situations” (p. 2).

Fifty adults were randomised to receive either delayed auditory feedback treatment either with transcranial direct current stimulation or with a sham treatment. The delayed auditory feedback treatment involved “six intervention sessions in consecutive days,” (p. 5), each being 20 minutes, but no further details were provided. Percent syllables stuttered was measured 1 week pre-treatment and immediately pre-treatment, and at 1 week and 6 weeks post-treatment. Measures were based on 2 minutes of within-clinic reading, monologue and conversation speech samples, which were of 550–630 syllables duration.

At 6 weeks post-treatment, the group that received the transcranial direct current stimulation reduced %SS scores by one third. The delayed auditory feedback treatment was inert: the group that received only that treatment showed no improvement of %SS scores at 6 weeks post-treatment. Perhaps controversially, the authors recommended the procedure for clinical use, stating that “brain stimulation using tDCS is not expensive and is simple to use” (p. 12). Arguably, though, more comprehensive speech measures are needed to endorse its clinical use; the report involved 2-minute speech samples, 6 weeks after the treatment, in the clinic, and under the speaking conditions in which the treatment was conducted.

## OTHER MACHINE-AIDED TREATMENTS

### Historical attempts

The history of clinical stuttering research contains many attempts to establish machine driven treatment.<sup>24</sup> Two such devices that could be worn by those who stutter were the electronic metronome and the Edinburgh Masker. Both these devices relied on two of the fluency enhancing conditions mentioned during Lecture One. The electric metronome was a device resembling a monaural hearing aid that produced a metronome beat to the ear. The Edinburgh Masker presented a masking signal during speech. This device involved a throat microphone held to the outside of the larynx with a strap, a headset, and a masking unit carried on a belt or in a pocket. In the long run, though, the value of these devices was never determined by a clinical trial, and they appear to be no longer clinically available.

### More recent attempts

#### *SpeechEasy*

This device incorporates altered auditory feedback (see Lecture One). It resembles a monaural hearing aid and presents altered auditory feedback to the wearer. A clinical trial that involved its commercial developers reported positive results,<sup>25</sup> but an independent Phase I trial<sup>26</sup> found no clinically significant effects, and the authors did not recommend moving on to a Phase II trial. This device has been the source of much controversy,<sup>27</sup> and it is not yet clear whether it is clinically useful.



#### *Modifying Phonation Intervals*

This device uses an ingenious idea that seems to be the opposite of speech restructuring. Rather than adding speech pattern features to control stuttering, it takes some away. What it takes away are short phonation intervals. It does this by means of a throat microphone held in place with a band, a signal processing box, and a display on a laptop computer. The laptop display indicates the proportions of phonation intervals within certain duration ranges and gives the speaker feedback on attempts to reduce the number of phonation intervals within certain ranges. The potential of the device is



supported by several laboratory research reports,<sup>28,29,30</sup> and a report<sup>31</sup> showing that four of seven clients who received a standard speech restructuring treatment reduced frequencies of short phonation intervals post-treatment. Two data based case studies were published.<sup>32,33</sup> A comprehensive clinical trial has yet to be published.

#### *Electromyographic (EMG) biofeedback*

An electromyography (EMG) machine uses surface electrodes to detect muscle action potential as muscles contract, and displays it visually. Clients attempt to change muscle action potential using such feedback. The procedure is used to treat several tension-related conditions and found its way into basic stuttering research with several promising laboratory reports.<sup>34,35,36,37,38</sup> A clinical trial involved EMG biofeedback<sup>39</sup> and reported positive results. However, two non-randomised trials failed to replicate those positive results.<sup>40,41</sup> No further research into this method has occurred.

## PHARMACOLOGICAL TREATMENTS

### *A long search*

Since the 1960s, there has been a search for a pharmacological stuttering treatment, and many clinical trials have been conducted for various compounds. These include anticonvulsant agents for treating epilepsy, antidepressants, antipsychotic agents, cardiovascular agents, and dopamine antagonists.

### *Two reviews*

A 2006 review of this topic<sup>42</sup> identified 31 reports that met what the authors determined to be minimum methodological requirements. The authors identified trials using the above mentioned pharmacological compounds and concluded that

The results of this systematic review of pharmacological treatments for stuttering are straightforward and are overwhelmingly negative. Of 31 studies reviewed, only 11 met three or more of five basic methodological criteria. Of those 11 plus 4 other relatively well-designed articles, only 1 provided data to show that stuttering was reduced to below 5%, the lenient outcome criterion selected for this review ... One other provided data to show that stuttering was reduced by at least half ... (p. 348)<sup>42</sup>

The authors concluded their review with a statement that there is no evidence that anyone who stutters has ever benefited from drug treatment and that it is unlikely that anyone ever will. Some other authorities in the field agreed with them.<sup>43</sup>

A more recent review<sup>44</sup> of drug trials for children and adolescents, extending back to the 1960s, included two older papers and one more recent paper that were not covered by the previous review. Seven papers were identified that met prescribed methodological criteria. Only one paper was found that the authors felt was methodologically strong enough to constitute reasonable evidence, and it found that a cardiovascular agent had no effect on stuttering. The authors concluded with the suggestion, that, surprisingly, never seems to have been thought of before: that drug treatment for stuttering might be improved if combined with standard speech-language pathology interventions.

A subsequent report<sup>45</sup> did evaluate the effects of olanzapine and haloperidol when added to a speech-language pathology treatment. However, the treatment was far from "standard," comprising "mixed treatment sessions that included 'air flow technique' and 'break Valsalva maneuver (sic.)' as well as 'desensitization' from Van Riper's protocol" (p. S271). This, plus the absence of contemporary speech measures, and side effects "such as mild drowsiness, dry mouth, and lethargy" make the results of the trial not compelling.

The search for a drug treatment continues, with a constant stream of preliminary reports. For example, there has been a preliminary study of ecopipam,<sup>46</sup> a dopamine receptor antagonist, with 10 adults who were studied during 8 weeks of using the drug. Stuttering severity reductions were measured from 900–2,300 syllables of within-clinic speech. Some stuttering reductions, and reductions of OASES scores, did occur, but arguably were not sufficiently large enough to be clinically significant. An

intriguing report contained a suggestion that green tea improved the speech and psychological status of adolescents who stuttered compared to controls.<sup>47</sup>

In short, at present there is no reason to consider drug treatment for stuttering. Regular reports continue to emerge about the effects of various drugs on stuttering, but they continue to not conform to accepted standards for a clinical trial in this field. An analysis of 114 social media posts<sup>48</sup> reported on the use of psychedelic drugs by adults who stutter. Not surprisingly, overall positive effects on stuttering were reported, but with side effects also reported. Another report called for clinical trials of such drugs.<sup>49</sup>

## SELF-HELP GROUP SUPPORT

As with many health issues, self-help support is available for those who stutter. Prominent examples are the National Stuttering Association in the United States, the Canadian Stuttering Association, and the Australian Speak Easy association. The Stuttering Association for the Young is a United States group for those younger than 18 years, with a branch in Australia. A systematic review of the effects of self-help support groups indicated general psychological benefits.<sup>50</sup> Facebook groups may be a useful alternative to in-person group support.<sup>51</sup> However, a report indicated that only around 1% of those who stutter engage with stuttering communities.<sup>52</sup> That estimate included social media, national stuttering associations and self-help groups.

Arguably, self-help activities for youth will be particularly useful. A report documented the benefits of a 2-week residential camp held by the Stuttering Association for the Young (Camp SAY) for 107 children who stuttered aged 8–18 years.<sup>53</sup> The camp is not formal therapy focused on stuttering. Its purpose is to promote “social interaction with people who stutter as a means of achieving positive change for the participants” with activities including “theater, swimming, arts and crafts, horseback riding, and sports” (p. 20). Campers have an opportunity to meet with a speech-language pathologist once or twice per week for additional support. OASES scores were collected on the first day of camp, the last day of camp, and at 6 months follow-up. There were small but statistically significant changes of OASES mean Impact Scores, which were sustained at follow-up. The changes may not have been clinically significant, with all means remaining within the mild-moderate impact score range.

Another camp-style 1-week residential program<sup>54</sup> contained components more directly targeting stuttering: communication and resiliency, mentorship and leadership, peer relationships, understanding bullying and teasing, and desensitisation about stuttering. For 23 children ages 4–14, evidence was presented to suggest that those activities reduced the impact of stuttering.

A 10-year follow-up of a 3-week intensive camp treatment in Germany involved 10 participants who were 9–20 years old at the time of the camp.<sup>55</sup> After the intensive component, “refresher meetings” occurred for 3–6 days, three times each year. The camp activities involved a combination of Van Riper’s techniques (see Lecture Eight) and speech restructuring. The authors concluded that “intensive stuttering therapy in childhood or adolescence can have a long-term positive affect on both internal and external stuttering symptoms” (p. 1). Naturally though, as the authors noted, the uncontrolled nature of such a design with so few participants requires a cautious interpretation.

As discussed during the next lecture, speech-related social anxiety is likely to be a clinically significant issue for children who stutter, and the effects of this style of intervention on that aspect of childhood stuttering require future documentation.

## CLINICAL NOTES ABOUT PERSISTENT STUTTERING: ADOLESCENTS

### A life transition

Clients in the age range 13–17 years are a unique group. The adolescent—teenage—years are a transition from childhood to adulthood and are accompanied by changes not experienced at any other time of life. Clinical challenges associated with adolescence arise from changes during this period that span physical, cognitive, emotional, and social domains. These changes can impact the client and the

family,<sup>56,57</sup> and can be a significant consideration when planning and implementing treatment.

Adolescents can be thought of clinically as neither children nor adults.<sup>58</sup> Many adolescents will experience stress at some period during this time of life, which needs to be taken account of clinically, but many will not.<sup>59,60</sup> Other common features of adolescence that may need to be accounted for clinically are the emergence of a sense of independence and autonomy,<sup>56,61,62</sup> developing importance of the peer group,<sup>59,63</sup> and a decline of motivation.<sup>64</sup> The emerging importance of the peer group is associated with a need to fit in to the norm,<sup>57</sup> which can cause problems if an adolescent stutters.

### Parents during treatment

A report of how 13–17 year old adolescents interacted with their parents about stuttering<sup>65</sup> is useful information for clinicians who treat this age group. An important finding was that some of these adolescents made the decision to attend a speech clinic themselves, and for others it was the parents who motivated them to do so. Most of the adolescents reported that they found parent assistance with their stuttering to be helpful, but it was clear that unsupportive parent input might occur. They commonly reported that helpful parent input

involved the use of speech skills, where parents would remind participants to use certain speech strategies, provide advice, or sometimes practice speech skills with participants. Also, participants reported parents providing assistance in the form of listening and not interrupting them when talking. (p. 50)<sup>65</sup>

However, the minority who reported unsupportive parent input found it to be “frustrating and perceived in an unsupportive manner” (p. 50).

### Telehealth and adolescents

For adolescents, there are advantages to telehealth. Telehealth treatment, which appears viable for adolescents,<sup>54</sup> can give them the independence that is important to them. If they wish, their parents need not be involved in treatment at all. Parents, too, can benefit from telehealth treatment of their adolescent children. For example, they do not need to take them to and from the clinic. Video telehealth presents treatment within an adolescent-friendly medium that facilitates client empowerment and self-management. Finally, the Internet for social purposes, using laptop, tablet and smartphone devices, are now part of the lives of most adolescents who have access to such technology.<sup>66</sup> Treatment methods for this age group can readily include technology<sup>67</sup> such as the Scenari-Aid website.<sup>68</sup>

A preliminary trial considered during the previous lecture,<sup>69</sup> which showed positive treatment outcomes, revealed some useful information for this treatment format with adolescents. Naturally, with so few participants conclusions must be guarded, but there was some suggestion that the video telehealth format was a better prospect for these adolescents than those in the other Phase I trial<sup>51</sup> who were treated in a clinic.

At another source describing this telehealth trial,<sup>70</sup> one adolescent participant commented that for treatment sessions she did not need to “race home from school and get ready and go somewhere” and found treatment “comfortable ... because I was in my own house and in a more familiar place.” Another said treatment was easier because he could “just hop up on my computer instead of going to the clinic.” His father said, “he was just so relaxed. I think this is a big call but he is probably more relaxed with [his SLP] than he is at the school he goes to, and with his teachers. This is a big winner.”



## CLINICAL NOTES ABOUT PERSISTENT STUTTERING: SCHOOL-AGE CHILDREN

### The evidence for changing tractability

Clinical trials of early intervention compared with clinical trials of adult intervention indicate that stuttering is at its most tractable shortly after onset and at its least tractable during adulthood. This seems to be related to neuroplasticity of the childhood brain (see Lecture Seven). Effect sizes are larger for early childhood stuttering than for adults, novel speech patterns are not required as part of efficacious clinical management, and there are far fewer signs of relapse than with adults. The adolescent clinical trials considered during this lecture provide no reason to believe that adolescent clients are more clinically tractable than adults. Hence, it seems possible that a change in tractability might occur during the primary school years.

A Phase III clinical trial of the Lidcombe Program for children with early stuttering<sup>71</sup> shows no sign that %SS outcomes are affected by age. Table 5 (p. 662) of this report shows that outcomes were the same for the 28 children in the trial younger than 4 years as for the 19 children older than 4 years. So, there was no sign of changing treatment responsiveness during the period of early childhood stuttering.

However, it seems to be a different story with the retrospective follow-up of children previously treated with the Lidcombe Program when they were 6–10 years old.<sup>1</sup> One of the children in the report, Participant 4, did not respond to the treatment, apparently because of compliance issues. For the remaining children, aged 6–10 years, in Table 2 (p. 284), analysis shows a significant, moderate negative correlation between age in months when treatment began and the percentage reduction of %SS scores at follow-up ( $r = -.72, p = .012$ ).<sup>†</sup> In other words, there is evidence that increasing age during the school years is associated with decreasing treatment effect sizes at follow-up. For the 11 children in this report, around half of the treatment effect at follow-up can be accounted for by age at the time of treatment ( $r^2 = .52$ ).

So, to summarise, verbal response contingent stimulation is suitable and efficacious for children with early stuttering, but speech restructuring is usually suitable for adults, and there is reason to believe that responsiveness to verbal response contingent stimulation decreases with age during the school years.

### A systematic review

A systematic review<sup>72</sup> explored treatments with potential to reduce stuttering in school-age children. The review considered clinical reports with an outcome involving any measure of stuttering for children 6–12 years old. The review established 67 studies with methods that involved data-based case studies, nonrandomised pre-treatment to post-treatment reports, and nonrandomised studies with a control group. The 67 studies suggested treatment effects of different sizes. The authors grouped the treatments as “Operant methods,” “Speech restructuring (including syllable-timed speech),” “Combined operant methods and speech restructuring,” “Machine-driven treatments” (self-modelling was included in this category) and “Treatments with a cognitive behaviour therapy component” (p. 6). The authors concluded that all available evidence was weak but, nonetheless, those treatments were suitable targets for future clinical trial development.

The authors of the review pointed out the advantages of operant treatment methods for this age group; they require no speech pattern, and the child does not risk any negative social effects from using an unnatural sounding speech pattern. They noted that operant methods show indications of the largest potential treatment effects. However, they noted the potential benefits of speech restructuring with this age group and drew attention to the potential combination of speech restructuring and operant methods. The authors pointed out some indications in the studies reviewed that combined speech restructuring and CBT components have particularly strong therapeutic benefits. A conclusion to the review was that several promising treatments have been overlooked for this age group and their

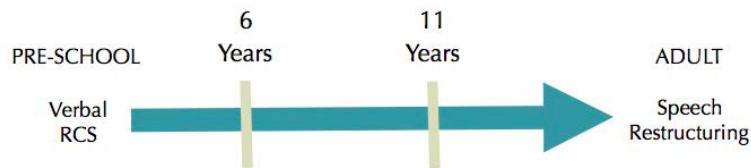
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<sup>†</sup> This analysis is not reported in the paper.

potential benefits have not been explored fully with clinical trials.

### A treatment selection model

The information above can be incorporated into a treatment selection model for school-age children. The model would, with increasing age, have verbal response contingent stimulation becoming progressively less suitable during that time of life, and speech restructuring becoming progressively more suitable. The model is presented in the figure.



The model suggests verbal response contingent stimulation as a first intervention of choice for school-age children, because it is a simple treatment and does not require a novel speech pattern. The last treatment of choice for school-age children would be the more complicated speech restructuring technique, with its associated disadvantages, supplemented with video self-modelling.

The potential viability of another supplement to speech restructuring for this age group was established with a laboratory study of 7–12-year-olds who stuttered.<sup>73</sup> Under dual-tasking conditions, where they spoke while conducting a non-linguistic task, their stuttering-like disfluencies decreased. The authors suggested that those preliminary findings have clinical promise.

Considering the clinical importance of the school-age time of life for stuttering, with its apparent changing clinical tractability, it is regrettable that clinical research to guide clinicians with this age group is so sparse. Reviewers of interventions for this age group<sup>74</sup> have not been able to identify a consistently efficacious treatment for them. The gravity of this situation prompted the editor of an international speech-language pathology journal dealing with school-age children to issue a call to rectify the situation urgently,<sup>75</sup> and more than 100 researchers and clinicians endorsed that call.<sup>76</sup>

### Adaptation of the Lidcombe Program for school-age children

The model of treatment selection for school-age children in the diagram above may prompt clinicians in some circumstances to use the Lidcombe Program with that age group, considering that there is encouraging clinical trial evidence of its value for them.

However, some adaptations to the clinical process are necessary when using the Lidcombe Program with school-age children. The language used to present verbal contingencies will be different, as perhaps will the activities for presenting verbal contingencies during practice sessions. It is also realistic to expect children to participate more actively in the treatment, such as engaging with parents and the clinician in scoring and recording SR scores and using self-imposed contingencies such as spontaneous self-correction. For a recent video telehealth clinical trial of the Lidcombe Program with 6–12-year-olds, techniques are explained for how to adapt the treatment for that age group, along with video demonstrations.<sup>77</sup>

### Teachers and school-age children who stutter

#### *The classroom*

Lectures Ten and Eleven deal with the common association between stuttering and social anxiety. Not surprisingly, then, there are several sources of anecdotal and research evidence that implicate fear of speaking in the classroom as a potential issue for stuttering primary school children.<sup>78</sup> A common clinical picture of a school-age stuttering child is one who is quiet and withdrawn, reluctant to participate in classroom activities, and is constantly anxious about being called on to speak in class.

As expressed in an early report:

In school, he generally sits in the rear of the class, rarely initiates discussion or answers questions spontaneously, and he avoids most situations which might provoke the slightest fear of stuttering. Even though he may be intellectually superior to most of his classmates, he minimizes his own potentialities, capacities, and gifts by remaining silent and not risking the possibility of a stuttering effect. (p. 141)<sup>79</sup>

That depiction is consistent with a more recent report of interviews with 30 adult stutterers.<sup>80</sup> All but one reported beginning to conceal stuttering during the school years, and seven reported doing so during or before elementary school before 11 years of age.



### *The importance of teachers*

That early report<sup>79</sup> noted the importance of early school experiences because they often represent the first excursion into the world by children without daily parent contact. Accordingly, teachers can be critical personnel in the lives of children who stutter, particularly during the school years. If a stuttering child is anxious in the classroom and feels that it is a dangerous and threatening place, a teacher can make the classroom feel much safer:

I very rarely put my hand up to answer a question because I didn't want to make a fool of myself. But the teachers generally understood me and I made good friends with a lot of my teachers.<sup>81</sup>

Participants who were interviewed as young adults made it clear that the importance of the classroom experience extends into adolescence.<sup>82</sup> An adaptation of the OASES (see Lecture Four) for carers (OASES-C) reported that teachers of children 2–6 years old associated stuttering with a mild-to-moderate impact.<sup>83</sup>

### *How teachers might help*

A useful teacher approach to fear of classroom speaking seems to have been originally suggested in 1940.<sup>84</sup> This paper contained sensible advice to teachers of not suggesting to children any techniques for controlling stuttering. Instead, the teacher can confidentially discuss with a child how help might be offered in the classroom, and together they can formulate a strategy for handling the matter. The interview study mentioned previously<sup>82</sup> indicated that such a constructive, individualised approach rarely occurred for the participants. A report of New Zealand teachers suggested that they have limited knowledge of strategies to assist students who stutter.<sup>85</sup>

It might be expected that anxiety about reading aloud in class can worsen for children who stutter when the class takes turns to speak. Apprehension about speaking, and quite often physiological signs of anxiety, can build steadily. Such anxiety about an impending classroom speech has featured in interviews of adults recounting school experiences:

If I thought there was a teacher that would randomly pick kids to read or would go down the row and everybody gets a turn, I'd have my mother talk to them and once again explain my situation, so that I did not have to read in class because any time they started that my ears would get hot, I'd start getting nervous, I couldn't sit still, I just started to sweat, and the only thing I could think about was counting down the time until I had to read. (p. 77)<sup>86</sup>

I used to go upstairs to the second floor bathroom and just keep flushing the toilet so that nobody could hear me getting sick, and then that kind of physical behavior lasted with me a long, long time because as a young adult and as an adult whenever I had a speaking situation coming up I would get physically ill. (p. 78)<sup>86</sup>

One student, they would read out a few stanzas. Then the next student would read out a few stanzas, and I could just tell immediately I was going to be number 22 there in the line. I just I couldn't think of anything else. And that moment of dread when my when it was my turn. And I hated that. I just hated that.<sup>81</sup>

An account by United States President Joe Biden is similar.<sup>87</sup> A common sense approach here would be for the teacher to ask the child for a preference about speaking order. For example, if the child wishes to speak toward the start of the order and has a name towards the end of the alphabet, the sequence of children speaking could occur in reverse. Or the teacher could call on children randomly, except for the child concerned, who is called on at an agreed time. Or, if the child is sitting towards the back or the front of the class, the speaking could be done in order of seating position.

A comprehensive review<sup>88</sup> examined the empirical evidence for common recommendations about how teachers can assist with school-age students who stutter. That review pointed out that many recommendations have been made to call on students to speak early during the class to reduce anxiety. The review points out that there is no direct empirical evidence about the merits of that approach. However, some laboratory experiments of wait-time to speaking with stuttering participants suggest that it is justified. Based on these experiments, the review presents the caveat that the student “should not be the initial speaker or should not read in the earliest position” (p. 9).<sup>88</sup>

The review describes two additional approaches to children who stutter in the classroom: anti-bullying interventions and giving a presentation about stuttering. A report<sup>89</sup> showed that an anti-bullying school program, involving 4 hours of teaching with manuals and videos, could positively influence peer attitudes and bullying for school-age students who stutter. Another report<sup>90</sup> suggested that a 45-minute presentation about stuttering improved attitudes about stuttering, although participants were adolescent students. A subsequent report found that improvements were retained 7 years later.<sup>91</sup> Positive results were reported with a 9-year-old boy who included a classroom presentation about his stuttering treatment,<sup>92</sup> and by speech-language pathologists who gave a classroom presentation as part of treatment for a 10-year-old girl.<sup>93</sup>

#### *Some modern resources*

Some modern suggestions about how teachers might help children who stutter in the classroom are available.<sup>94</sup> A video production by The Michael Palin Centre in London promotes teacher awareness of stuttering,<sup>95</sup> and is available at their website.<sup>94</sup> This video is a useful resource for clinicians who have contact with teachers of children who stutter. Clinicians may also direct parents to it so they can show it to the teacher of their stuttering school-age child. An overview of how speech-language pathologists might assist school-age clients with bullying includes teacher involvement.<sup>96</sup> A comprehensive stuttering resource has been developed in New Zealand.<sup>97,98</sup>

## **SUMMARY AND CONCLUDING COMMENTS**

The school-age years are a critical time for stuttering intervention. This is because of some remaining neuroplasticity of the childhood brain at that time of life, and also because of the social challenges imposed during the school years. The well-known effects of social anxiety with stuttering, as described in the following lectures, may occur in the classroom with 7–12-year-olds.

The most encouraging findings for stuttering treatment in school-age children come from non-randomised Phase II clinical trials of the Lidcombe Program. One caveat to those findings is that the Lidcombe Program treatment model was developed for young children, and so it will not be suitable for all school-age children. Guidance is available for adapting the treatment for that age group.<sup>77</sup> A trial<sup>4</sup> showed that a third of school-age children attained Stage 2 criteria, and some clinical benefits occurred for the other children. Overall, the Lidcombe Program is not as effective for school-age children as with pre-schoolers. However, the obvious clinical advantage of a verbal response contingent stimulation treatment model for school-age children is that it does not require a speech pattern. Consequently, post-treatment speech naturalness probably will not be a clinical issue. There is some evidence (the Oakville Program) that combining verbal response contingent stimulation with

syllable-timed speech has clinical benefits.

For school-age children, a systematic review<sup>72</sup> identified some promise in a model of programmed verbal response contingent stimulation. This is known as Gradual Increase in Length and Complexity of Utterance, commonly referred to as GILCU.<sup>99,100,101</sup> However, there has been only one report by the developers of the treatment that qualifies as a Phase I clinical trial according to the Lecture Five definition.<sup>102</sup> Modest results were reported, and the treatment seems to have attracted no clinical or research interest lately. Also during the last century, a response contingent stimulation method of self-imposed time-out was developed.<sup>103</sup> When a stuttering moment occurs, the speaker stops for a few moments, then resumes speaking. Phase I and II trials have been reported this century,<sup>104,105</sup> but, as with GILCU, this operant treatment seems to have attracted no clinical or research interest lately.

At present there is no reason to consider either pharmacological treatment or machine-aided treatment for any client. However, according to some researchers, there is future potential with non-invasive brain stimulation.



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## LECTURE TEN: STUTTERING, SOCIAL ANXIETY, AND MENTAL HEALTH<sup>†</sup>

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### BACKGROUND

#### A changing view about stuttering and anxiety

##### *Past decades*

Research perspectives about stuttering and anxiety have changed during recent decades. A review of the topic at the close of the 20<sup>th</sup> century<sup>1</sup> showed that, during the mid 1980s, prominent scholars of the day agreed that there was little convincing evidence of a relationship between stuttering and anxiety.<sup>2,3,4</sup>

However, a follow-up review of the area 10 years later<sup>5</sup> showed that things had changed. Continued research, with methodological improvements, allowed the authors of the review to conclude that there is compelling evidence of a relationship between stuttering and anxiety. The authors of the review drew attention to mounting evidence of clinically significant anxiety levels associated with stuttering, and evidence that adults who stutter may—but not necessarily—experience psychological problems related to anxiety. They reported progress in clinical management of anxiety with those who stutter, and new ways to measure it clinically.

##### *More recent views*

A 2019 survey of 502 adult stutterers<sup>6</sup> indicated the presence of negative, anxiety-related emotions. Around half of them reported “often or always” experiencing embarrassment (53%), feeling emotionally drained (49%), and feeling ashamed (45%) during moments of stuttering (p. 4339). In 2021, there was evidence of research in the field detecting anxiety early in the lives of those who stutter. A systematic review and meta-analysis of the topic<sup>7</sup> concluded that “the summary effect size indicates that children and adolescents who stutter present with increased anxiety symptoms ... compared with non-stuttering peers” (p. 1).

Accordingly, research about stuttering and anxiety reviewed in this section has profoundly influenced modern clinical practices. In 2021, the views of 12 scholars in the field<sup>8</sup> included the need for core clinical assessment covering domains related to speech anxiety: “(d) reactions to stuttering by the speaker; (e) reactions to stuttering by people in the speaker’s environment; and (f) adverse impact caused by stuttering” (p. 2379).

### ANXIETY

#### Expecting harm

Generally speaking, expectancy of harm drives anxiety. Examples of exceptions to this generalisation include innate infant anxiety states, such as fear of separation and fear of strangers. It appears that anxiety involved with stuttering focuses on anticipation of harm in social situations, where speech is required. Examples of such harm would be social rejection, being laughed at, or being ignored. In fact, one report showed expectation of such social harm to specifically be the issue; those who stutter appeared to be troubled only by such expectations, not expectations of any physical harm.<sup>9</sup>

#### Three components

Anxiety is commonly described as a complex psychological state composed of verbal-cognitive, behavioural, and physiological components.<sup>1,10,11,12</sup> Clinical psychologists group the following cluster of emotions as related to anxiety: scared, shy, panicky, and insecure.

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<sup>†</sup> Thanks to Ross Menzies and Lisa Iverach for guidance with this material.

*Verbal-cognitive*

Anxiety commonly includes thoughts and expectancies about negative, harmful events. The prevailing psychological perspective is that, except for some evolutionary anxiety responses, such as fear of heights, water, and spiders, emotions come from thoughts that emerge in response to events.

The kind of harm that might be expected by those who stutter in social situations is fear of negative evaluation. They may perceive social situations and performance based situations, such as addressing a group of people, as threatening and dangerous. They may have a fear that listeners in such situations will form a negative view about them, and that negative view will cause them harm in some way, such as humiliating or demeaning them.

Naturally, most people will experience social anxiety in some situations, particularly when speaking to a group of people. However, at some point, anxiety about such situations becomes clinically problematic because it interferes with usual enjoyment of life activities. The destructive and unhelpful thoughts that can drive the social anxiety of those who stutter are well known and discussed shortly. Examples include thoughts that “people will wonder what’s wrong with me if I stutter” and “people will think I’m strange.”

*Behavioural*

People who are socially anxious may avoid social situations to some extent, or to use the common expression, they may be socially avoidant. They may also avoid situations that focus on speech performance, such as talking to a group of people. Or, in extreme instances, they may show escape behaviours once they are in such situations by leaving them.

*Physiological*

Those who stutter and are anxious about social situations may endure them with considerable distress, which could, but not necessarily, be manifested with physiological symptoms. They may experience symptoms such as sweating, blushing, increased heart rate, heart palpitations, hyperventilation, dry mouth, shortness of breath, nausea, headache, shaking, and muscular tension. Those physiological symptoms can prompt cognitive symptoms such as mental blocking, difficulty concentrating, and feeling flushed. During research, physiological anxiety responses can be measured with salivary cortisol, skin conductance, blood pressure, respiration rate, and heart rate.

*Loosely connected components*

Clinical psychologists think of the above three anxiety components—verbal-cognitive, behavioural, and physiological—as being loosely related to each other.<sup>11</sup> They do not systematically increase or decrease together in a lawful way. From one anxiety-provoking situation to another, one of them may increase while another may decrease or not change. They will not necessarily all be present when someone is anxious. This feature of anxiety is well known in clinical psychology and has been shown to be the case with those who stutter.<sup>13,14</sup> The latter of those reports involved participants speaking to a virtual audience and a virtual empty room. Although a measure of the verbal-cognitive anxiety component (Subjective Units of Distress; see Lecture Eleven) was elevated in the former condition, stuttering severity, heart rate, and skin conductance showed no changes.

One study of 13 adult stutterers and 15 controls<sup>15</sup> showed elevated skin conductance and lower respiratory sinus arrhythmia in the stuttering group. Respiratory sinus arrhythmia is heart rate fluctuations linked to breathing that occur naturally; during inspiration, heart rate increases and during expiration heart rate decreases. Lower respiratory sinus arrhythmia is associated with social anxiety. One of two self-reports of anxiety in the study showed an association with respiratory sinus arrhythmia.

Clients who stutter may be distressed by destructive thoughts and beliefs about negative social evaluation but will not necessarily have any signs or history of situation avoidance, escape, or physiological arousal. It is quite possible for someone who stutters to always enter feared situations, and experience distress when doing so, but without any physiological signs. Probably, with most clients who stutter, the prominent sign of anxiety is what they will tell clinicians: thoughts about



harmful social evaluation.

#### *State and trait anxiety*

Anxiety linked to personality, or temperament, is referred to as *trait anxiety*. In contrast, the term *state anxiety* can be used to refer to immediate emotional responses to everyday experiences. Naturally, there is a link between the two, with those having trait anxiety likely to experience more state anxiety than others.

## STUTTERING, ANXIETY, AND ANXIETY DISORDERS

### Stuttering and anxiety

#### *Last century*

There is an extensive research literature, with much recent growth, showing that those who stutter are more socially anxious than controls. Indirectly, this is verified by research, overviewed in Lecture Three, that more stuttering occurs with larger and more threatening audiences. Key findings from the last century were that those who stutter have high levels of anxiety according to standard clinical psychology measures,<sup>16,17</sup> and that stuttering and control subjects can be accurately distinguished using such measures without any speech data.<sup>18</sup> That report developed the Speech Situation Checklist<sup>18</sup> for the identification of speech-related anxiety with participants who stuttered and participants who did not. The checklist refers to situations such as “talking to a stranger,” “being interviewed for a job” and “introducing yourself,” and asked participants to rate how each situation caused “fear, tension, anxiety, or other unpleasant feelings” (p. 354).<sup>18</sup> For 21 selected situations, the reported strength of such emotions was able to predict with 93% accuracy whether participants stuttered or not. This result was replicated in a later study<sup>19</sup> comparing treatment-seeking adults with controls. Results showed that 10 of the Speech Situation Checklist responses were 97% accurate for distinguishing between the groups. A survey that included 621 Australian adult stutterers<sup>20</sup> asked about “anxiety level due to stuttering.” Around a quarter indicated that they were “a little anxious,” around a quarter indicated that they were “fairly anxious,” and around a quarter indicated “very anxious.” There were 12% who indicated “extremely anxious,” and 10% who indicated “not anxious” (p. 6).

Salivary cortisol is a physiological marker of anxiety which has been associated with stuttering several times.<sup>21,22</sup> A study of 19 adult men who stuttered and 19 matched controls<sup>23</sup> reported higher levels in the former group. Dry mouth (xerostomia) can be a symptom of anxiety, and the stuttering participants showed a higher score than controls on a short self-report measure known as the Xerostomia Inventory. Consistent with this result, a physiological measure of unstimulated saliva flow rate was lower for the stuttering participants than the controls. Physiological markers of skin conductance and heart rate, together with self-report psychological measures, have found to be elevated with stuttering participants compared with controls.<sup>24</sup>

#### *This century*

Research findings this century continue to confirm that, as a group, adults who stutter are socially anxious compared with those who do not stutter.<sup>12,25</sup> A meta-analysis<sup>26</sup> confirmed this for trait anxiety and social anxiety. Studies were included in the analysis if they used established psychometric measures of trait and social anxiety, and if they compared a stuttering group of participants with a control group. Eleven studies dealt with trait anxiety and eight dealt with social anxiety. Findings confirmed that those who stutter differed as a group from those who do not stutter, with an effect size (see Lecture Five) of  $d=0.57$  for trait anxiety and a much greater effect size of  $d=0.82$  for social anxiety. The latter effect size showed those who stutter to be nearly a standard deviation above controls for social anxiety measures. In short, any individual who presents to a clinic with stuttering will likely—but by no means certainly—have a history of social anxiety. There is some evidence that anxiety-related problems with stuttering may affect racial and ethnic groups differentially.<sup>27</sup> The United States National Health Interview Surveys for 2010–2015, based on 875 parent reports for 4–17-year-olds, suggest that Hispanic and African American children were less likely than Caucasian children to

show early signs of such problems.

### *Responses to anxiety and related emotions*

A survey study<sup>28</sup> that included 180 adult stutterers generated evidence that ways of coping with anxiety and related negative emotions about stuttering affect the impact of stuttering measured with the OASES (see Lecture Four). Emotional regulation refers to methods that can be used to deal with emotional states. The Emotion Regulation Questionnaire<sup>29</sup> is a self-report measure with 10 items to measure cognitive reappraisal and expressive suppression. Cognitive reappraisal involves changing emotions by changing thinking about situations that causes them.<sup>†</sup> Expressive suppression is a less constructive approach that involves dealing with emotions by not expressing them. For adults, the survey study<sup>28</sup> found that less frequent use of cognitive reappraisal and more frequent use of expressive suppression were associated with higher OASES impact scores. Although significant, the associations accounted only for 12% and 14% of OASES scores.

## **Stuttering and social anxiety disorder**

### *What is social anxiety disorder?*

*Social anxiety disorder*, once known as *social phobia*, is described in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5)<sup>30</sup> published by the American Psychiatric Association. Social anxiety disorder involves a pervasive fear of humiliation and embarrassment in social and performance situations. Those affected have an intense fear of negative evaluation and judgement by others.<sup>31</sup> Social anxiety disorder can have adverse effects on the lives of those affected, causing social avoidance, and generally restricting the usual enjoyment of interactions with others. For example, it produces fear and avoidance of activities such as disagreeing with others in a social situation, expressing a controversial viewpoint, and in any way being the centre of attention.<sup>32</sup> Social and performance situations are commonly avoided, or endured with extreme distress, often accompanied by the physiological symptoms described earlier. An important feature of social anxiety disorder is that, considering the actual threat, the expectation of humiliation and embarrassment in social situations is unrealistic and irrational.

### *Stuttering and social anxiety disorder*

There is evidence that those who stutter and seek clinical help are likely to have social anxiety disorder. The population prevalence of social anxiety disorder is 8–13%.<sup>33,34</sup> However, case reports of social anxiety disorder are common for those who stutter,<sup>35,36</sup> with the condition reported for 40%,<sup>37</sup> 44%<sup>38</sup> and 60%<sup>39</sup> of cases in speech clinics. The latter of these reports indicated that such cases have 34-fold increased odds of meeting criteria for social anxiety disorder diagnosis compared with age and gender matched community controls. A study of 84 Japanese school students,<sup>40</sup> with a mean age of 14.6 years, reported that 50% of them met diagnostic criteria for social anxiety disorder. These reports are consistent with studies reporting that, in general, adults who stutter have anxiety scores higher than controls but slightly lower than those with psychiatric conditions.<sup>17,41,42</sup> However, a report of older stuttering participants after a lifetime with it<sup>43</sup> did show anxiety scores in the range associated with social anxiety disorder. A review of social anxiety disorder and stuttering is available.<sup>44</sup> A report<sup>45</sup> compared the demographics of clients presenting to speech clinics for stuttering treatment with and without social anxiety disorder. Apart from the group with social anxiety disorder being significantly younger, no demographic differences were found.

As outlined during Lecture One, a stuttering moment can involve repeated movements, fixed postures, or superfluous behaviours. It is intuitive to predict that the presence of social anxiety in those who stutter might be connected to stuttering that is more behaviourally complex, containing stuttering behaviours that are potentially more socially distracting, with more fixed postures and superfluous behaviours than repeated movements. However, there is evidence that is not the case. A study of 3,100 stuttering moments from 147 adolescents and adults<sup>46</sup> showed no relation between the

<sup>†</sup> This approach is used in cognitive behaviour therapy, which is described in Lecture Twelve.

behavioural complexity of stuttering and the presence of mental health disorders, anxiety, or depression. Based on that result, the researchers indicated that, clinically, there is no reason to expect that the presence of complex stuttering behaviours suggests concomitant mental health issues.

#### *The reality of peer responses to stuttering*

There is no doubt that many, if not most, adults with clinically significant stuttering have experienced negative peer social reactions at some stage because of their stuttering.

Oh, over the years I've seen various responses to my stuttering where people would laugh, people would avoid eye contact, things like that, or they would smile. But everyone reacts to it in a slightly different way. It is something I've just got used to over the years.<sup>47</sup>

However, for a social anxiety disorder diagnosis, the expectation of social humiliation and embarrassment must be unrealistic and irrational in relation to the actual threat.

I think when I'm actually stuttering, I feel as if people are responding inappropriately sometimes. But when I stand back and objectively and objectively assess the situation, most people don't really care that much. So I think it's important not to take things too personally.

I will say what really annoys me is my inability to, I suppose, properly communicate like others can communicate. And it makes me feel slightly inadequate at times. But I think at the end of the day, it's important to realize that while stuttering is a barrier to communication, it isn't a too big of a barrier and that I can communicate well even with a stutter.<sup>47</sup>

Arguably then, the prevalence of social anxiety disorder and stuttering is consistent with the results of a study involving 324 adults from the United States<sup>48</sup> that was mentioned during Lecture Two. The study involved a list of 15 stigmatising experiences, and "most participants reported experiencing them never or rarely in the past year" (p. 55).<sup>48</sup> The list included experiences such as "people have been unkind to me," "people have avoided looking at me," and "people made fun of me or picked on me" (p. 55). Yet, most participants agreed that "because of my stuttering, I have worried about other people's attitudes toward me," "I am fearful that others will reject me if they hear me stutter," and "because of my stuttering, I have felt embarrassed in social situations" (p. 56). Not surprisingly, then, a report<sup>49</sup> showed that adults who stutter perceive their communication competence to be below that of controls.

It is of interest, then, that there is no research that directly measures exactly how often those who stutter encounter negative social peer reactions during everyday life. One report from 1954<sup>50</sup> showed that many "store clerks," when spoken to by someone who was stuttering, reported that they experienced embarrassment, pity, and sympathy. There were some early reports of physiological listener responses to stuttering, and a more recent report<sup>51</sup> verifies such negative reactions and provides evidence also of listener skin conductance and heart rate changes when confronted by severe stuttering. There are data<sup>52,53,54</sup> that show listeners looking away from videos of people stuttering more often than for control speech. A report<sup>55</sup> suggested that, overall, listeners do not respond differently in terms of turn-taking behaviours when talking with those who stutter and those who do not. Findings showed, though, that during stuttered utterances, conversational partners tended to interrupt and complete the utterance for the speaker. Also, for those with moderate compared with mild stuttering, listeners used more reinforcers such as "um-hum" and "right." However, all this does not quantify how often such peer reactions occur during everyday life. Nor does it indicate whether someone who stutters would necessarily be aware of such reactions.

## **Stuttering and other anxiety-related disorders**

### *DSM-5 disorders*

Many DSM-5 psychological disorders involve anxiety. Some examples are generalised anxiety disorder, mood disorders, depression, and personality disorders. There is some evidence that who

stutter are at risk of having these disorders also.

There are reports<sup>56,57</sup> that show stuttering clients who seek treatment when compared to controls have 4.5 times more chance of having generalised anxiety disorder, 2.1 times more chance of having any mood disorder, 1.9 times more chance of experiencing a major depression, and 3.0 times the chance of having any of the personality disorders, three of which are related to anxiety: obsessive compulsive disorder, dependent personality disorder, and avoidant personality disorder. However, another report<sup>58</sup> failed to find any evidence of personality disorders among adults seeking treatment for stuttering.

#### *The Symptom Checklist-Revised*

A study of 200 stuttering and control adults<sup>59</sup> with the Symptom Checklist-Revised (SCL-90-R)<sup>60</sup> found significantly elevated scores for the former group, with many symptoms that may involve anxiety: somatisation, obsessive compulsiveness, interpersonal sensitivity, depressive and anxious mood, hostility, phobic anxiety, paranoid ideation, and psychoticism. A subsequent report from that cohort<sup>61</sup> reported the relationship between the Global Severity Index, which is an overall measure of mood state from the Symptom Checklist-Revised, and a measure of self-efficacy, which is the expectation of being able to accomplish tasks. Findings were that, for a 5-month period, positive self-efficacy was associated with better mood. Another report<sup>62</sup> found that 30–40% of 129 adults who stutter experienced negative mood, and it was noted that the rate was similar to that of social anxiety disorder.

#### *Substance abuse*

There is a strong association between anxiety and substance abuse. However, this seems not to be the case with those who stutter and seek treatment.<sup>57</sup> That finding was replicated with a community sample within a British birth cohort study<sup>63</sup> where participants who reported no stuttering at 16 years (N= 10,491) were compared with those who did report stuttering (N=188). There was no evidence of an association of stuttering with alcohol or smoking.

#### *Sleep patterns*

A report from a United States health data base<sup>64</sup> studied sleep patterns in adult stutterers. The authors' motivation for the study was a suggestion about irregular biological rhythms in the literature about temperament and early childhood stuttering,<sup>65,66</sup> and two reports<sup>67,68</sup> from the United States National Health Interview Survey linking insomnia with childhood stuttering. The report used the National Longitudinal Survey of Adolescent Health (N=13,564). Part of the survey collected data about insomnia and hours of sleep. Participants who indicated "yes" to the question "do you have a problem with stuttering or stammering?" (p. 4) (n=261) reported an average of 20 minutes less sleep per night than controls. Additionally, those participants with self-reported stuttering were twice as likely to have insomnia than controls. The authors interpreted their findings in terms of a link between sleep problems and anxiety, and hence between sleep problems and stuttering. The same research group documented self-reported sleep apnoea symptoms among the same group of participants who reported stuttering.<sup>69</sup> Compared to controls, they reported significantly more difficulty concentrating and more fatigue during the day. The same group replicated these findings later,<sup>70</sup> and they were replicated with Iranian participants.<sup>71</sup> The finding was also replicated with 50 Brazilian stuttering children 6–12 years old<sup>72</sup> and matched controls. Parent questionnaire data showed that 21 of the stuttering children were above the cut-off for sleep problems, compared to only two controls.

#### *Depressive symptoms and suicidal ideation*

Another report used the National Longitudinal Survey of Adolescent Health (N=13,564) to explore depressive symptoms and suicide ideation.<sup>73</sup> Respondents were asked questions about depressive symptoms and whether they had "seriously considered committing suicide in the last 12 months" (p. 4). Results showed that, compared to controls, adolescents and young men and women who stuttered reported higher levels of depressive symptoms. For the females, there were signs of depression increasing with age. The males who stuttered (but not the females) were more likely than controls to report suicide ideation. However, another report<sup>74</sup> followed up a United Kingdom cohort (N=3,824)

and found that stuttering at 8 years of age did not predict self-harm with suicidal intent. There was a report that teenage girls who stutter experience more negative impact on their lives than teenage boys.<sup>75</sup>

A study of suicide ideation<sup>76</sup> involved 140 adults with self-reported stuttering, 76% of whom reported receiving treatment, and 27% of whom reported being clinically diagnosed with anxiety or depression. All participants completed a screening for suicide risk, and 50% of them scored at the positive screening cut-off for suicide ideation. The authors stated that these values are above population rates for suicide ideation. Many participants reported that their suicidal thoughts were related to their stuttering, and scores for suicide ideation were reported to be associated with OASES-A scores. The authors provided appropriate caveats for interpreting their data. The most important of these are that most of the sample reported receiving treatment for stuttering, and that the sample may have been biased also because potential participants were aware that the topic of the study was suicide ideation. For the latter reason, the sample may have over-reported the population incidence of suicide ideation. The authors also pointed out the need for inclusion of specialised health services in cases of stuttering clients who have considered self-harm.

## ANXIETY IMPAIRS SPEECH TREATMENT

### The problem of post-treatment relapse

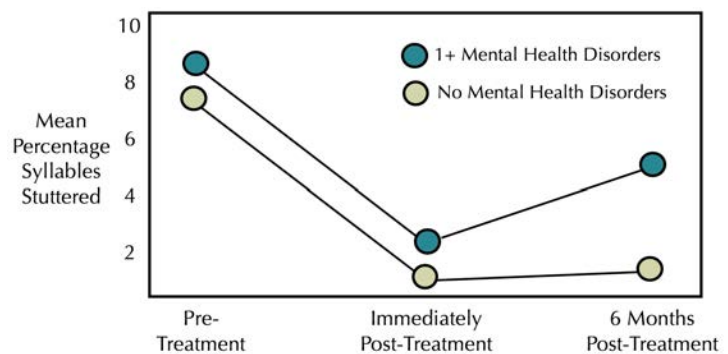
For decades it has been known that, after speech restructuring treatment to reduce or eliminate stuttering, only around one third of clients can sustain their treatment benefits.<sup>77,78</sup> In other words, the relapse rate after speech treatment is around two-thirds:

One-third of the clients achieved and maintained satisfactory fluency ... one-third of the clients achieved satisfactory fluency during treatment but experienced significant regression over time ... almost one-third of all clients studied either failed to complete a treatment program or were unavailable for subsequent follow-up assessment. (p. 16)<sup>77</sup>

### Anxiety and post-treatment relapse

The first reported association between anxiety and treatment outcome seems to have been in 1976.<sup>79</sup> A subsequent publication showed that the one-third of clients with self-reported relapse had elevated anxiety scores on the Spielberger Trait Anxiety Scale.<sup>78</sup> This was followed with a study of 64 adults who received a speech restructuring treatment in clinical trials.<sup>80</sup> Around two-thirds of them were diagnosed with having one or more mental health disorders, the majority of which involved anxiety. Everything looked fine for the one-third of the group who had no such mental health disorders, as shown in the figure.\* Immediately after treatment the stuttering severity of the group reduced and remained that way 6 months later. This is a classic example of a successful short-term speech treatment outcome.

But consider the two thirds of the group who had one or more mental health disorders. This is a much different result. In the first instance, those clients had more severe stuttering than those who had no mental health disorders. They also were a less clinically responsive group. They reduced



\* Adapted and reproduced with permission: Iverach, L., et al. (2009). The relationship between mental health disorders and treatment outcome among adults who stutter. *Journal of Fluency Disorders*, 34, 29–43. © 2009 Elsevier.

their stuttering by around two thirds, but the group with no mental health disorders reduced their stuttering by nearly 90%. However, the important finding is that at 6 months post-treatment the group with no mental health disorders showed little sign of relapse. But this is not at all the case for the group with one or more mental health disorders, who started to relapse. This is a classic example of a poor short-term treatment outcome because of relapse.

## THE CLARK AND WELLS MODEL OF SOCIAL ANXIETY DISORDER

### The puzzle

Those who have phobias such as heights, water, and spiders, generally manage to avoid what they fear and never learn that their assessment of the threat is unrealistic. For example, those who are flight phobic may never fly. Consequently, they may never learn that what they fear—that the plane will crash—does not happen. However, for social anxiety disorder, it is a different matter. For those affected, even if they are socially avoidant in general, it is virtually impossible to avoid social encounters entirely. Yet they still don't learn that their constant and pervasive fear of social humiliation and embarrassment is unrealistic. In other words, they persist with that belief even though experience provides constant evidence to disconfirm it.

### The Clark and Wells model

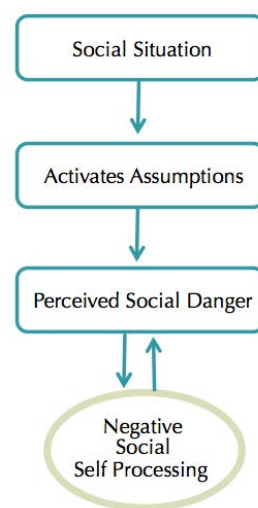
A major contribution of this model<sup>31</sup> is that it explains that puzzle about social anxiety disorder; why social anxiety persists in the face of constant experiences that should disconfirm the belief that social situations are harmful. There are many models to explain this,<sup>81</sup> and one has been devised specifically for stuttering.<sup>82</sup> However, the Clark and Wells model is the most influential, having been confirmed several times with tests of hypotheses derived from it. The Clark and Wells model seems to dominate clinical programs for social phobia internationally. Psychologists commonly incorporate the model of social anxiety disorder within their social anxiety management procedures, with efficacious results.<sup>83</sup> Components of the model feature in a treatment for the social anxiety of those who stutter that will be discussed during the next lecture. An overview of the model has been presented in an easily understandable way,<sup>84</sup> and this overview is essential reading for speech-language pathologists who commonly encounter stuttering clients with social anxiety. The following description of the model draws freely from that source.

### Three assumptions of the model

The model rests on three assumptions. The first involves the existence of excessively high standards of social performance, such as believing you should always be entertaining and intelligent, and must never make a social slip-up: "I must not show any signs of weakness" and "I should always have something interesting to say" (p. 406).<sup>84</sup> The second assumption involves beliefs about performing in a certain way in social situations: "If I am quiet, people will think I am boring," "If people get to know me, they won't like me" (p. 406).<sup>84</sup> The third assumption of the Clark and Wells model is unconditional negative self-beliefs, such as "I'm boring," "I'm stupid," and "I'm different from everyone else" (p. 407).<sup>84</sup> Such assumptions cause a perception of an impending social situation to be threatening, as shown in the figure.\*

#### *Negative self-processing in social situations*

An impending social situation activates the assumptions just described, as shown in the figure, which signal that the impending social situation is threatening, making it a source of perceived social danger.



\* Adapted and reproduced with permission: Clark, D. M. (2001). A cognitive perspective on social phobia. In W. R. Crozier & L. E. Alden (Eds), *International handbook of social anxiety: Concepts, research and interventions relating to the self and shyness* (p. 405–430), Hoboken, NJ: Wiley. © 2001 John Wiley & Sons, Ltd.

Then, during the social encounter, negative self-processing prevents disconfirmation of the social event as dangerous.

### *Observer perspective*

A prominent aspect of that negative self-processing is that attention shifts from the actual situation that is occurring towards an image or impression of what people think is occurring: how they think they appear to others. One way this is expressed is that the person affected sees an *observer perspective* of the situation rather than a *field perspective*. When most people are asked to recall a social encounter, they will relate to their own field of vision—their field perspective—which is of course what is appropriate for them to recall.

However, those affected with social anxiety disorder, and other anxiety disorders also, are known to report an observer perspective.<sup>85</sup> They will report how they looked to others in the social situation, which is usually uncomfortable and awkward, from the perspective of an observer. It is obvious that something is amiss with this, because it is impossible to see an observer perspective of yourself; it is only possible to imagine one. This prevents disconfirmation of perceived danger from the actual social situation: “what they see in the image is not what the observer would see but rather their fears visualized” (p. 408).<sup>84</sup>

Findings about observer perspective have been reported with interviews of adult stuttering and control participants.<sup>86</sup> The stuttering group reported more recollection of intrusive and recurrent mental imagery than controls. The stuttering group were distinctive for their recurring imagery themes of shame, sadness, helplessness, and frustration.

Another report<sup>87</sup> of 30 participants seeking treatment for stuttering and 30 controls involved recall of a situation in which they felt anxious. The stuttering group was significantly more likely than the control group to recall impressions and images that were from the observer perspective. Further, the stuttering group was more likely to recall images that were negative, distorted and from that observer perspective. The authors concluded that the results could be caused by the same maladaptive social processing that occurs with anxiety disorders. This is an example of observer perspective recall from a stuttering participant:

It was kind of over the shoulder, over my right shoulder. You could just make out the side of my head and the top of my shoulder and the person I was talking to was sort of in the full frame. And um occasionally when I was having trouble saying something I would kind of turn my face to the right so you could get a profile look of my face and how I’m struggling to complete the sentence. (p. 5)<sup>87</sup>

A survey of 502 adult stutterers<sup>6</sup> showed signs of observer perspective. The response option “often or always” was selected by 17% of participants for “mentally check out, that is, I feel removed or like I’m watching from afar” (p. 4339).

### *Bias toward negative social stimuli*

The other aspect of negative self-processing is that those with social anxiety are disinclined to recognise positive social input that would disconfirm the threatening nature of social encounters. For example, they avoid positive faces in favour of negative faces,<sup>88</sup> detect negative social information more accurately than positive social information,<sup>89</sup> have slow recognition of positive social stimuli,<sup>90</sup> and pay excessive attention to emotional social stimuli.<sup>91,92</sup>

There has been a finding to this effect with stuttering participants,<sup>93</sup> who looked at positive faces less often than controls when speaking to a group. They also looked less often and for a shorter time at all audience members when compared with controls. The results of Stroop task<sup>†</sup> studies<sup>94,95</sup> are broadly consistent with that finding. The latter of these two studies reported that, compared with controls, participants in the stuttering group showed bias toward socially threatening words such as “inept,”

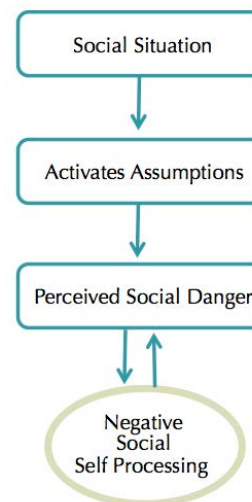
<sup>†</sup> With Stroop tasks, participants name the colours of the text in which different words are printed. The task can be used to assess reaction time interference.

“foolish,” “failure,” and “inadequate.”

Another method that has shown negative attentional bias with socially anxious participants is the dot-probe task, which is sometimes referred to as the probe detection task. A study of 43 adolescents who stuttered and 43 controls<sup>96</sup> with a dot-probe task found that the stuttering group had attentional bias to threatening faces. Another dot-probe study with fewer participants, 12 adult stutterers and 14 controls,<sup>97</sup> used a “social threat induction task.” Participants were told they would give a 5-minute talk about a topic, to be announced at short notice, which would be recorded and shown to a group of public speaking experts. After being exposed to that social threat, compared to controls, the participants who stuttered showed an attentional bias toward negative facial expressions.

One report has failed to show such an effect for stuttering participants, at least for those who were not socially anxious.<sup>98</sup> Another report<sup>99</sup> with 48 adult stutterers and 42 controls failed to find any negative bias in responses to written descriptions of various social situations.

So, if someone is socially anxious and perceives social situations as dangerous, a destructive cycle of negative thought processing can begin during the social encounter, and over subsequent encounters, those thoughts make the perceived threat worse. Overall, this intensifies the negative self-processing. That destructive cycle is shown with the bidirectional arrows in the figure.\*



## Safety behaviours

*Attempting to prevent feared outcome*

Safety behaviours are used by those who are socially anxious as an attempt to prevent a perceived threat or negative event from occurring. Those with social anxiety disorder use them commonly in social situations. Examples of safety behaviours commonly used by those with social anxiety disorder are to reduce the chance of social penalty by avoiding eye contact, allowing a conversational partner to do most of the talking, and keeping answers short. A list of common safety behaviours for the general population is presented in the Subtle Avoidance Frequency Questionnaire.<sup>100</sup> For the safety behaviours used by those who stutter, a checklist is available.<sup>101,102,103</sup>

*Safety behaviours prevent fear extinction*

There is evidence<sup>104</sup> that safety behaviours in fact maintain anxiety by preventing learning that fears are unfounded, and that situations are not as dangerous as they are perceived to be. Or, to use the correct jargon, safety behaviours prevent *fear extinction*. So, avoiding eye contact stops someone learning that people are giving accepting looks, and gives the mistaken impression that avoiding eye contact provides protection from harm, when in fact there was no potential harm. Likewise, allowing a partner to do most of the talking prevents learning that nothing socially negative happens from talking.

*Safety behaviours can cause the feared outcome*

In addition to preventing fear extinction, safety behaviours can have the effect of causing feared social outcomes to occur, rather than providing protection from them. For example, speaking little, keeping answers short, and avoiding eye contact, can make someone appear to be uninterested in engaging with others. This can lead to the feared negative outcome. Accordingly, with the Clark and Wells model of social anxiety disorder, the use of safety behaviours in response to the original perceived

\* Adapted and reproduced with permission: Clark, D. M. (2001). A cognitive perspective on social phobia. In W. R. Crozier & L. E. Alden (Eds), *International handbook of social anxiety: Concepts, research and interventions relating to the self and shyness* (p. 405–430), Hoboken, NJ: Wiley. © 2001 John Wiley & Sons, Ltd.



social danger can feed into a cycle of worsening perceived social danger, as shown in the figure. \*

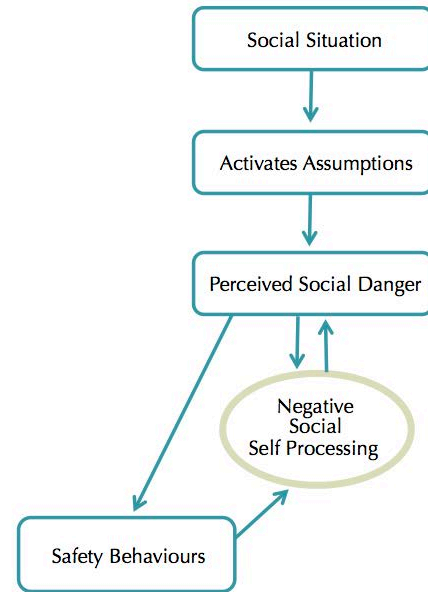
*Eliminating safety behaviours during anxiety treatment*

Safety behaviours are routinely targeted for elimination during anxiety treatment.<sup>31,84,105</sup> A review<sup>106</sup> of studies of anxiety-related conditions, such as social anxiety disorder and obsessive-compulsive disorder, showed that removal of safety behaviours promoted reduced perception of threat in the long term.

*Safety behaviours during stuttering treatment*

It appears that health professionals can unwittingly promote safety behaviours through problem solving advice for their clients.<sup>107</sup> There is evidence that this occurs with speech-language pathologists who treat stuttering clients.<sup>101</sup> That report surveyed 160 speech-language pathologists and reported evidence that they may recommend what might, in effect, be safety behaviours to adult clients in order to manage anxiety. The survey results generated a list of 34 potential safety behaviours, and factor analysis revealed five categories of them. They are presented in the table below.

The most recommended of the *General Safety Behaviours* was avoiding anxiety-provoking topics. Clinical psychologists recognise this strategy as a common safety behaviour that is intended to protect against social threat. Clinical psychologists also recognise silent rehearsal before speaking as a safety behaviour, and most of the participants reported recommending it to clients as a *Practice and Rehearsal* safety behaviour. More than half of the speech-language pathologists reported giving advice listed under *General Avoidance* once or more.



SAFETY BEHAVIOUR CATEGORY	EXAMPLES
<i>General Safety Behaviours</i>	<i>Avoid topics that make you anxious</i> <i>Ask many questions</i> <i>Point rather than speaking</i> <i>Allow your partner to talk for you</i> <i>Talk little</i>
<i>Practice and Rehearsal</i>	<i>Rehearse mentally before speaking</i> <i>Practice the speech restructuring technique just prior to speaking</i> <i>Rehearse answering the phone mentally before answering</i>
<i>General Avoidance</i>	<i>Avoid unnecessary talking on a bad day</i> <i>Keep answers short</i> <i>If anxious avoid difficult words</i>
<i>Choose Safe and Easy people</i>	<i>Immediately before an important speaking situation</i>

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	<i>In socially threatening situations</i>
<i>Control Related</i>	<i>Say "relax" to yourself when anxious</i>
	<i>Speak slowly when anxious</i>
	<i>Try to take deep breaths</i>

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The authors of the study concluded that further research is needed to determine how often clients follow such advice. Additionally, they stated a need to determine whether any, or all, of such recommendations are in effect safety behaviours if clients do use them. In other words, there is a need to determine whether they are adaptive and helpful, or in fact prevent fear extinction.

Indeed, in clinical psychology it is not currently clear which client behaviours should be considered safety behaviours and which behaviours should be considered healthy adaptive behaviours in a situation that causes anxiety.<sup>104,107</sup> For those who stutter, an interesting perspective about this matter emerged from a survey of 122 adults.<sup>108</sup> Self-reported word and situation avoidance was associated with impaired quality of interactions with health care providers.

A subsequent report<sup>102</sup> of 133 clients who sought anxiety treatment for stuttering indicated that 132 of them reported using one or more of 27 safety behaviours. Most reported were:

*"try to avoid difficult words," "rehearse sentences mentally before saying them," "keep your answers short," "choose safe or easy people to talk to in socially threatening situations," and "try to avoid difficult syllables." (p. 1249)*

There was evidence that the reported use of many of the safety behaviours correlated with scores for measures known to be associated with stuttering-related anxiety: Fear of Negative Evaluation and Unhelpful Thoughts and Beliefs About Stuttering (see Lecture Eleven). This suggests a connection between anxiety and use of safety behaviours by those who seek clinical help for stuttering.

A survey of 502 adult stutters<sup>6</sup> showed signs of safety behaviour use with their selection of the response option "often or always" for these survey items: "remain silent and choose not to speak" (15% of participants), "remove myself from a situation" (10% of participants), and "let someone else speak for me" (9% of participants) (p. 4339).<sup>†</sup>

#### *Safety behaviours and speech restructuring*

A review of this topic<sup>109</sup> drew attention to how clinical use of speech restructuring could place clients in a situation where they attain control of stuttering at the expense of perpetuating speech-related anxiety. In other words, there is a potential conflict between speech restructuring and management of social anxiety:

For those clients who wish to control their stuttering and where speech restructuring is deemed the most suitable approach, it is possible that speech restructuring may (a) induce or increase self-focused attention, (b) promote the use of safety behaviors, and (c) become a safety behavior itself. (p. 59)<sup>109</sup>

The authors of that article present a detailed approach to dealing with this clinical challenge.

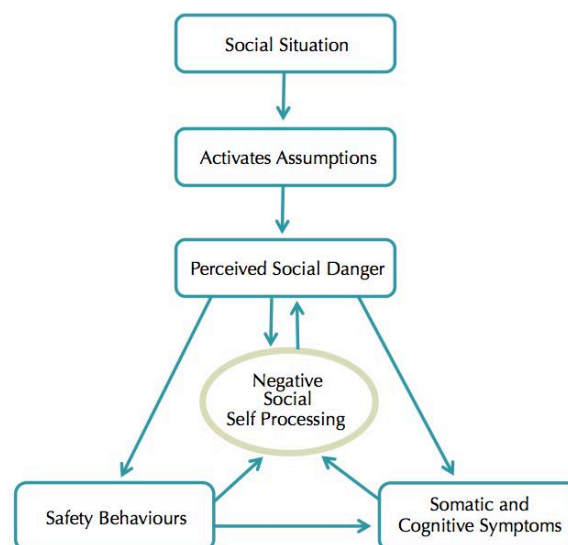
#### *Self-disclosure and safety behaviours*

In this context, it is worth considering the commonly recommended technique of self-disclosure with clients who stutter. The technique was overviewed during Lecture One. As noted during that lecture, recommendations for its use connect it with speaker anxiety. In which case, it may be a safety behaviour. The technique may, in fact, prevent fear extinction and have the effect of exacerbating a feared outcome.

<sup>†</sup> This report describes these safety behaviours as covert stuttering.

## Somatic and cognitive anxiety symptoms

The other part of social anxiety disorder is that anxiety is not necessarily confined to unhelpful thoughts, as discussed previously. As soon as anxiety is present somatic symptoms could occur, as shown in the figure:\* sweating, blushing, increased heart rate, heart palpitations, hyperventilation, shortness of breath, nausea, headache, shaking, feeling flushed, and muscular tension. These physiological symptoms may prompt cognitive symptoms such as mental blocking and difficulty concentrating. The person then becomes self-focussed and attends to these physiological symptoms as well as cognitive appraisals such as negative thoughts. This process acts to confirm negative thoughts and beliefs that the situation is dangerous. Such responses can feed into a destructive cycle associated with social encounters to make the whole experience extremely distressing, not only for psychological reasons but for physical reasons.



### Before the feared situation

The Clark and Wells model deals not only with what occurs during the social situation but what happens before and after it. It is typical for those with social anxiety disorder to ruminate in advance of the situation about all the past failures and negative social experiences that they have had. This can even occur so vividly that the start of the negative self-processing that occurs within a situation may even occur before it happens. This recollection of past failure, which is not at all based on reality, can lead to so much expectation of a repeat episode that the person may at this point choose to avoid the situation rather than enduring the distress of being in it. Again, all this provides further failure to disconfirm irrational beliefs about social dangers.

### After the feared situation

After the event, those with social anxiety disorder can conduct a “post mortem of the event” (p. 411).<sup>84</sup> Rumination may continue in a destructive fashion, even though the anxiety and distress associated with the event might have subsided. In fact, such rumination might reaffirm a belief that the event was negative, and it is added to a list of past failures. Post-mortem rumination about innocuous events can be interpreted as a reflection of poor self-worth, such this example:

a patient at a dinner buffet mentioned how much he liked a bread and butter pudding. Later in the evening, he heard his hostess say she disliked bread and butter pudding. Afterwards, he thought his comment revealed he was unsophisticated and worthless. (p. 411)<sup>84</sup>

A study of post-event rumination with 96 adults who stuttered<sup>110</sup> involved a socially stressful situation of giving a 5-minute, impromptu speech to a group of strangers. Results showed that self-rated stuttering severity, OASES scores, and social anxiety were strong predictors of scores on a test<sup>111</sup> of post-event rumination.

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**SUMMARY**

Anxiety is a psychological state composed of loosely connected verbal-cognitive, behavioural, and physiological components. Adults with stuttering who present at speech clinics often will have clinically significant anxiety that requires intervention. Many such cases will require referral to a clinical psychologist. Such adult clients may warrant a DSM-5 diagnosis, notably social anxiety disorder. If an adult does have clinically significant anxiety, it reduces the chance of effective speech treatment. The Clark and Wells model of social anxiety disorder is applicable to stuttering. Speech-language pathologists need to be mindful of the possibility that techniques for stuttering control may be safety behaviours that sustain speech-related anxiety.

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## LECTURE ELEVEN: THE ORIGINS OF SOCIAL ANXIETY WITH STUTTERING

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### THE ORIGINS OF SOCIAL ANXIETY WITH STUTTERING: THE EARLY YEARS

Obviously, the social anxiety problems that commonly trouble adults who stutter begin at some time earlier in life. It is essential, then, when young children present to clinics with stuttering, to identify any signs that might signal the potential for future development of social anxiety problems.

#### Psychometric evidence

##### *The ELVS Cohort*

The report for the ELVS cohort (see Lecture Two) at 4 years of age<sup>1</sup> presented data from the Pediatric Quality of Life Inventory Parent-Proxy Report (PedsQL),<sup>2</sup> which is a medically oriented quality of life scale completed by parents. The ELVS cohort showed that, at 4 years of age, no differences for “psychosocial health related quality of life” (p. 464)<sup>1</sup> were associated with stuttering. There are qualifications about interpretation of those data because the PedsQL scale is not a standard measure of childhood anxiety used in clinical psychology literature. However, some of the scale items clearly do pertain to anxiety, such as “I feel afraid,” “I feel sad,” and “I worry about what will happen to me” (p. 139).<sup>2</sup>

The ELVS report<sup>1</sup> also featured the Strengths and Difficulties Questionnaire, which is an assessment used commonly in child mental health research.<sup>3,4,5</sup> It measures emotional and behavioural problems with five scales: “emotional symptoms, conduct problems, hyperactivity-inattention, peer problems, and prosocial behavior” (p. 1337).<sup>4</sup> There are 25 test items with five items for each of those scales. To each of 25 statements about the child, parents indicate either “not true,” “somewhat true,” or “certainly true.” A “total difficulties score” is obtained from the sum of all scales except the prosocial behavior scale. An “internalizing scale” is obtained from the sum of the emotional symptoms and the peer problems scales, and an “externalizing scale” is obtained from the sum of the conduct problems and the hyperactivity-inattention scales.

At 4 years of age in the ELVS report, no differences were reported for Strengths and Difficulties Questionnaire measures for the control children and the children who had begun to stutter.

##### *The Millenium Cohort*

However, another report<sup>6</sup> provided opposite results for the Strengths and Difficulties Questionnaire using the Millenium Cohort.<sup>7</sup> That cohort comprises some 19,000 children who were born in the United Kingdom during 2000 and 2001. One of the many questions asked of parents when their children were 3, 5 and 11 years old was whether the child had “stuttering or stammering” during the previous 12 months. At these ages parents were also asked to complete the Strengths and Difficulties Questionnaire, along with many other assessments. Data were available for 3-year-olds (n=173), 5-year-olds (n=194), and 11-year-olds (n=194) who were reported to be stuttering. The Total Difficulties scores showed statistically and clinically significant differences from controls at all ages. The report concluded that “cohort members who were reported to stutter were more likely than those with typically developing speech to experience behavioural, emotional and social difficulties” (p. 27) and that “early social, emotional and behavioural difficulties may be apparent in children who stutter as young as 3 years old” (p. 30).<sup>6</sup>

##### *The United States National Health Interview Survey*

The results from the Millenium Cohort were replicated in a study<sup>8</sup> that used data from the United States National Health Interview Survey for 2010–2015. Among many other health related questions, parents were asked whether their children had “stuttering or stammering” during the previous 12

months. A short, six-item version of the Strengths and Difficulties Questionnaire was used in the National Health Interview Survey.<sup>9</sup> The first five items “related to being well-behaved, experiencing worry, being unhappy or depressed, social behaviors, and attention to tasks,” and required parents to respond, “not true,” “somewhat true,” or “certainly true.” The sixth item dealt with “functional impairment due to difficulties with emotions, concentration, behavior, or being able to get along with other people.” For this item, parents responded “no,” “yes,” “minor difficulties,” “yes, definite difficulties,” or “yes, severe difficulties.” Comparisons were made between children in the sample 4–5 years old whose parents reported stuttering (n=144) and whose parents did not report stuttering (n=7,171). Significant differences were found for all domains, with odds ratios around 2–3.

#### *The Generation R Study cohort*

A report<sup>10</sup> used the Generation R Study, which is a prospective Netherlands community cohort study from foetal life onwards. When the children were 9 years old parents were asked by survey “does your child currently stutter?” and “has your child ever stuttered in the past?” (p. 4566). Children were classified as “stuttering history” if one answer was “yes” and “stuttering persistence” if both answers were “yes.” The sample obtained was 3,421 children, of which 118 were classified as “stuttering history” and 27 were classified as “stuttering persistence.” To screen behaviour-related mental health problems in the children, the Child Behavior Checklist<sup>11</sup> was administered at ages 1.5 years, 4.0 years, 5.0 years, and 9.0 years. The children’s temperaments were assessed at 0.5 years and 6.0 years with the Revised Infant Behavior Questionnaire<sup>12</sup> and the Child Behavior Questionnaire.<sup>13</sup>

The study was built around two hypotheses: Hypothesis 1 was that behaviour problems and temperament during the pre-school years (1.5 years and 3.0 years) predict stuttering history and stuttering persistence. Hypothesis 2 was that stuttering history and stuttering persistence predict behaviour problems and temperament. Hypothesis 1 suggests causal involvement of behaviour problems and temperament in stuttering, and Hypothesis 2 suggests them to be an effect of stuttering. The authors concluded “we found hardly any evidence for our first hypothesis” (p. 4572). The only significant result from six Child Behavior Checklist and six Infant Behavior Questionnaire subtests was a temperament score of “recovery from distress.” There was support for Hypothesis 2: stuttering persistence during the pre-school years was associated with Child Behavior Checklist scores later in life, and with stuttering history associated with negative affectivity temperament scores at 6 years.

#### *Clinical cohorts*

A report of 427 children<sup>14</sup> included the Child Behaviour Checklist, the Short Temperament Scale for Toddlers,<sup>15</sup> and the Short Temperament Scale for Children.<sup>16</sup> The children were treated for early stuttering beginning when they were younger than 6 years. For that cohort of children who presented clinically, the authors reported “nothing unusual about behavioural and emotional functioning, or the temperaments” (p. 622) of the children. The report used the Depression Anxiety and Stress Scales<sup>17</sup> with parents and reported that their scores were within typical limits. However, the Recent Life Changes Questionnaire<sup>18</sup> suggested that “a third of parents were experiencing moderate to high life stressors at the time of seeking treatment” (p. 622). This led the authors to speculate that “life stresses were instrumental in prompting parents to bring their stuttering pre-schoolers to clinics, possibly with the onset of their children’s stuttering exacerbating such stresses” (p. 629).

Another report<sup>19</sup> was consistent with the ELVS report, showing no health related quality of life issues with a group of 197 children, ages 3–6 years, who were participants in a clinical trial of early stuttering treatment. Compared to normative data, there were no systematic problems for the children according to four medically oriented quality of life instruments.

#### *Small studies*

A report<sup>20</sup> incorporated data for eight children who showed pre-treatment scores in the typical range for the Child Behaviour Checklist. Another report<sup>21</sup> presented data for seven stuttering and seven control children and showed that they did not differ on the Preschool Anxiety Scale,<sup>22</sup> which is a more direct parent report measure of anxiety. This report also showed no differences for salivary cortisol. Those two reports, however, do not contain enough participants to be particularly convincing.

The Pictorial Scale of Perceived Competence and Social Acceptance for Young Children<sup>23</sup> is a direct assessment based on child responses. It was given to 28 children with stuttering, mean age 4 years 9 months, and a control group of children.<sup>24</sup> There were no significant differences between the two groups for test scores; however stuttering severity was a significant predictor of social acceptance, accounting for 20% of the variance in test results.

A significant result was found in a study of child stutterers and controls.<sup>25</sup> The report included 15 child stutterers and 15 controls aged 3–6 years. Significant differences were found for the Child Behaviour Checklist between the groups for all seven scales of the test, notably “Anxious Depressed” and “Emotionally Reactive.”

### Autonomic nervous system evidence

#### *Positive findings*

The physiology of anxiety and early childhood stuttering has been studied by comparing the autonomic nervous systems of children with early stuttering and controls.<sup>39,26</sup> One report<sup>39</sup> studied 20 children with early stuttering and 20 controls. The study dealt with the temperament feature of emotional regulation, using indices of sympathetic and parasympathetic nervous system activity.<sup>†</sup> Skin conductance was used to measure sympathetic activity, and respiratory sinus arrhythmia was used as a parasympathetic measure. Respiratory sinus arrhythmia is heart rate fluctuations linked to breathing that occur naturally; during inspiration heart rate increases and during expiration heart rate decreases. Lower respiratory sinus arrhythmia is associated with social anxiety in adults.

The children watched a neutral screen during a baseline condition.<sup>39</sup> Then they watched short videos that successfully elicited positive and negative emotions. Then, the children told a story about the videos. Some important significant differences between the two groups were reported. The child stutterers showed lower (parasympathetic) respiratory sinus arrhythmia during the baseline, which theoretically means they had increased vulnerability to a sympathetic response. Additionally, the child stutterers showed more (sympathetic) skin conductance increase during positive emotions while watching and talking about videos. Interestingly, a similar effect size was reported (Table 3) which was consistent with the observations in a previous review.<sup>38</sup>

The same research group used similar research methods to study parasympathetic activity of children 3–4 years old who stuttered (n=10), who had recovered from stuttering (n=200), and control children (n=36).<sup>27</sup> On this occasion, no baseline differences between groups was reported. Some “nuanced” (p. 11) but significant differences between the groups were reported for the children’s narratives after they viewed video clips that contained positive and negative emotional content.

The authors concluded from the earlier study<sup>39</sup> that their results suggested autonomic nervous system involvement with early childhood stuttering. They speculated about the nature of that involvement: that such activity may divert the necessary attentional resources from speech and communication. Naturally, the presence of unusual temperament and early childhood stuttering raises the caveat discussed earlier of whether such unusual temperament is involved in the cause of stuttering, or whether it is merely an effect of stuttering. The authors acknowledged this issue but argued that it is unlikely that their results were caused exclusively by the experience of stuttering.

In their conclusions to another study of respiratory sinus arrhythmia and early childhood stuttering,<sup>26</sup> those authors again presented the cause-or-effect caveat but argued a view that on balance, “cognitive, emotional, and related processes appear to play a meaningful role in the onset and/or developmental trajectory of childhood stuttering” (p. 2146). The same research group reported a skin conductance study of nine young children with persisting stuttering and 23 who they considered had recovered around 2 years later.<sup>28</sup> During a stressful picture naming task, data showed a significant 14% difference between the groups, but no differences from controls. Again, the authors concluded

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<sup>†</sup> The sympathetic nervous system controls responses to perceived threat, and the parasympathetic nervous system controls homeostasis at rest.

that “that emotion should be considered in any comprehensive account of childhood stuttering,” regardless of “the directionality of effect” (p. 149).<sup>28</sup>

A report by the same group<sup>29</sup> involved 18 child stutterers and 18 controls, mean age 4 years 5 months. The groups did not differ for baseline skin conductance, heart rate, or respiratory sinus arrhythmia. However, when presented with faces of negative valence, compared to controls the stuttering children showed an emotionally reactive effect in terms of higher heart rate and greater decrease of respiratory sinus arrhythmia. The authors concluded that “emotional reactivity and regulation has clinical significance for preschool-age stuttering and should be considered during assessment and treatment of stuttering in children” (p. 14), but no elaboration of that statement was provided.

#### *Two failures to replicate*

A study<sup>30</sup> of 47 child stutterers and 25 control children, with a mean age of 4 years 8 months, failed to replicate the findings discussed above. The children engaged in speech and nonspeech tasks, while measures were made of skin conductance and blood pulse volume. The researchers reported that “overall, the results of our study do not support the hypothesis that atypically high levels of sympathetic arousal are associated with speech production in preschool children who are stuttering” (p. 11).<sup>30</sup> Additionally, no differences were found between the two groups of children for a parent-reported temperament measure, the Children’s Behavior Questionnaire–Short Form, and the KiddyCat measure of children’s attitude to communication.

Another report<sup>31</sup> did not support the broad notion that there is autonomic system involvement with early childhood stuttering. Thirty-two children with early stuttering (mean age 3 years 11 months) and 16 controls (mean age 4 years 1 month) engaged in picture description and a more challenging nonword repetition task. Electrodermal activity was the same for both groups with the picture description task but was elevated for the nonword repetition task. For the group of child stutterers, KiddyCat scores were significantly higher, as were the Fear Scale and the Sadness Scale of the Children’s Behavior Questionnaire–Short Form. The researchers concluded that

Our findings suggest that age-appropriate social communication tasks are not inherently more stressful for preschool-age CWS and are not associated with state-related stress or anxiety that is often reported for adults who stutter. However, speaking tasks that place a higher demand on children’s cognitive–linguistic system may be more taxing and challenging to preschool CWS than CWNS, leading to a higher level of arousal. (p. 4030)<sup>31</sup>

### **Early childhood temperament**

#### *Temperament*

It is estimated that 20–60% of adult personality traits can be accounted for by temperament. Temperament is a stable, innate, and constitutional tendency to react to the environment or interact with it in a certain fashion. According to a popular definition,<sup>32</sup> “temperament describes our early emotional, motor, and attentional equipment, along with the regulating capacities that allow us to control our reactions and put them to good use” (p. 7). Temperament is influenced by biological features that include genetics. Childhood temperament is generally accepted as a risk factor for anxiety later in life.

The most used classification of childhood temperament was developed by Thomas and Chess,<sup>33</sup> and involves nine parent-reported dimensions. One of those is “approach/withdrawal,” which refers to how children respond to new situations: whether they readily engage in them or retreat from them. This dimension of temperament—behavioural inhibition or shyness—is regarded as a risk factor for anxiety disorders later in life.

#### *Temperament and early childhood stuttering: Some fundamental caveats*

There is a rapidly accumulating, diverse body of literature dealing with the association between early childhood temperament and stuttering. At present, this body of literature is inconclusive, and is being interpreted differently by many researchers. A reason for its inconclusive nature could be that it is

constrained by inherent limitations. So, a discussion of those limitations is useful before considering that literature.

A fundamental (and often overlooked) caution about this body of literature is that any findings of association between temperament and early childhood stuttering may pertain to speech and language disorders generally rather than to stuttering specifically.<sup>34,35,36</sup> That aside, given that temperament is a stable, innate construct present at birth, there is a potentially misleading implication in reports of temperament measured after the onset of stuttering. It might be more appropriate to think of “temperament” measures after stuttering onset as personality measures, which reflect the interaction between temperament and environmental experiences.<sup>†</sup> Indeed, inspection of the commonly used measures of early childhood temperament suggests that, in part, they reflect experiences of stuttering. In short, any association between early childhood stuttering and measures of temperament may reflect the effects of stuttering rather than its causes.<sup>37,38</sup> This clearly is a possibility because most children involved with research about stuttering and temperament have been stuttering for some years.

Another reservation about this literature is that it is observational, not experimental. In other words, it is based on simply observing the temperaments of children who stutter and children who do not stutter. As such, it can only establish an association between stuttering and temperament. It cannot definitively establish anything about the causality of stuttering.

And finally, many measures of temperament for children with early stuttering are based on parent report, which may be influenced by the presence of stuttering. In other words, when parents report about their children’s temperament characteristics, it is not clear to what extent their reports are confounded by the fact that their child stutters.

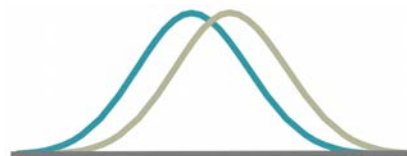
### *Three reviews of early childhood temperament and stuttering*

A full review of the topic is beyond the scope of this lecture, but the pertinent research is cited in three current reviews.<sup>36,37,38</sup> One of them<sup>36</sup> cites evidence that, for all children, a so-called “difficult” temperament heightens the risk of anxiety disorders later in life. “Difficult” temperament includes “nervous, high strung or tense,” “appears fearful or anxious,” “appears worried,” “not as happy as other children,” and “has difficulty having fun” (p. 153).<sup>36</sup> The review concluded:

Using the guideline that independent replication of findings makes them trustworthy, there is an inevitable conclusion to this review. For stuttering children during the preschool years, there may be some association between temperament and stuttering ... The guarded nature of this statement arises because of the modest scope of the research on which it is based, amounting to 10 publications, and because of some inconsistencies with results ... (p. 158)<sup>36</sup>

Another review<sup>38</sup> was published the following year and incorporated six new studies. Those authors also were tentative in their conclusion that “childhood stuttering may be associated with constitutionally based temperamental/emotional processes, many of which are believed to be open to environmental influences” (p. 128).<sup>38</sup> In another publication,<sup>39</sup> that research group summarised their view of the consistent findings to have emerged from a generally inconsistent literature about stuttering and early temperament. Compared with controls, they stated that children who stutter are (1) less adaptable, (2) have poorer attention and attention regulation, and (3) have a negative mood.

A review of the topic<sup>37</sup> focused on the effect sizes reported in the literature, recording all of them in tables. A central argument in the review is not only that inconsistent effects are reported but that maximum effect sizes reported are moderate. The review author makes the point that the two distributions for such an effect size look something like the figure. There is extreme overlap between the two groups and no real separation between them. All this led the reviewer to conclude:



<sup>†</sup> Thanks to Ross Menzies for this content.



Children who develop stuttering (as a group) are *not [author's italics]* characterized by temperamental traits such as shyness, social anxiety, or general anxiety ... A subgroup of CWS [*children who stutter*] tends to show somewhat elevated traits of inattention and hyperactivity-impulsivity. (p. 18)<sup>37</sup>

#### *Parent measures of temperament*

The prospective ELVS cohort<sup>40</sup> was used to determine the presence of temperament anxiety markers prior to and after the development of stuttering. This report involved 183 children with stuttering and 1,261 nonstuttering children, aged 2–4 years. At the children's second, third, and fourth birthdays, parents completed the Short Temperament Scale,<sup>41</sup> which is based on the Thomas and Chess temperament classification.

No differences were found at any age for the "approach" and "easy difficult" scales, which are thought to be anxiety precursors. At 3 years of age significant differences were found for the "reactivity" and "persistence" scales, indicating that at that age child stutterers "were less reactive to environmental stimuli and had a reduced ability to attend to a task until completion" (p. 1314).<sup>40</sup> However, there was no evidence for a continued difference with "persistence" at 4 years, and "reactivity" was not measured at that age. The authors concluded that there were

no signs of temperament precursors of anxiety before stuttering onset or shortly after. Results suggest, at most, that temperament is influenced somehow during the period after stuttering onset, but with a waning developmental influence subsequently. (p. 1314)<sup>40</sup>

A study of 123 children with ages 9–14 years<sup>42</sup> explored how temperament affects the impact of stuttering. Correlations were established between OASES scores and the Early Adolescent Temperament Questionnaire-Revised,<sup>43</sup> which is a self- and parent-report measure. Moderate correlations were found between "Surgency" and "Negative affect" temperament scores and OASES Overall Impact scores, which prompted a conclusion that

... on the one hand, more extravert and less fearful/shy children experience a lower overall impact of their stuttering; on the other, children with higher levels of irritability and frustration experience a higher overall impact of their stuttering. (p. 427)<sup>42</sup>

However, the authors noted that correlation does not establish causality, and that the study did not exclude the possibility that temperament scores simply reflect experiences with stuttering. Evidence to support that contention was reviewed earlier from the Generation R Study cohort.<sup>10</sup>

That conclusion from the Generation R cohort was consistent with the conclusion from the review noted earlier,<sup>40</sup> that temperament is most influential on early childhood stuttering but less so later in childhood. The authors of the study of 123 children<sup>42</sup> noted that a correlation between stuttering severity and the temperament dimension of "effortful control" had been reported for early childhood stuttering.<sup>44,45</sup> However, their study found no correlation between stuttering severity and temperament scores for 9–14-year-olds, and such a correlation seems absent also with persistent stuttering in adulthood.<sup>46</sup>

An innovative feature of the study of 123 children<sup>42</sup> was that, using the Early Adolescent Temperament Questionnaire, parent measures of temperament were supplemented with child measures. This is of interest because of the caveat mentioned earlier that parents' reports of temperament might be confounded by their children's stuttering. Moderate correlations were found between parent and child temperament scores, leading the authors to dismiss that prospect. However, because moderate correlations were found, another possibility is that parent reports about their children's temperaments might be influenced considerably by variables other than temperament, which might include the children's stuttering.

Using the same cohort of children, another report by the authors with 132 child stutterers, ages 9–14 years,<sup>47</sup> reported data from the Early Adolescent Temperament Questionnaire-Revised, and the Revised Children's Anxiety and Depression Scale.<sup>48</sup> The latter scale measures anxiety and depression

symptoms, with a parent- and a child-reported version. They reported significant, low to moderate correlations between child- and parent-rated versions of the temperament scale (surgency and negative affect) and the depression scale. However, the children's scores on the anxiety and depression scale were not considered to be clinically significant.

The second Illinois cohort<sup>49</sup> (see Lecture Two) studied 58 children with early stuttering and 40 control children. According to one subtest of the Children's Behavior Questionnaire-Short Form,<sup>50</sup> there was a significant difference between the groups during the 4–5 years of the study for Negative Affectivity. The children in the stuttering group who did not recover had greater negative affect than the control group and the children who recovered.

If there is a relationship between early childhood stuttering and temperament, there might be a correlation between stuttering severity and temperament measures. Several studies have reported such a correlation or association<sup>44,45,51</sup> and several have failed to show a correlation.<sup>42,47,52</sup> One of these reports<sup>53</sup> with 47 young children showed small but significant correlations between "percentage of stuttered disfluencies" during short narratives in a laboratory and "surgency," which is an index of emotional reactivity involving high levels of positive affect, derived from the Children's Behavior Questionnaire-Short Form. However, only around 10% of the variance was explained. The report failed to find any similar correlations for skin conductance.

Another study<sup>45</sup> followed up on an earlier finding<sup>44</sup> with 98 children from a university clinic, having a mean age of 6 years 7 months. The authors sought to replicate the earlier finding of an association between the temperament dimension of "effortful control" and parent measures of stuttering severity. The effortful control temperament dimension, measured with the Children's Behavior Questionnaire-Short Form,<sup>54</sup> describes the capacity to regulate focus and attention. A regression model found a statistically significant relationship between parent measures of stuttering severity and effortful control, with 13.6% of the variance explained.

### *Clinical applications*

There have been two suggestions<sup>39,44</sup> that an association between temperament and stuttering severity would contribute to sustaining early childhood stuttering. This idea, that early childhood temperament somehow contributes to the development of early childhood stuttering, has been echoed by other researchers.<sup>55</sup> Some have gone as far as suggesting the following about their findings:

Holistic treatment for stuttering in children should include the behavioral components of enhanced self-regulation of EC [*effortful control*] (i.e., attention, inhibitory control, and perceptual sensitivity) alongside traditional approaches to augment the effectiveness of each. (p. 12)<sup>45</sup>

Indeed, one of the authors took up this notion with a clinical trial, which will be discussed at the end of this lecture.

Some authors have gone as far as suggesting (cautiously) that there may be overarching clinical applications of findings associating temperament and early childhood stuttering.<sup>38</sup> In essence, they suggest that childhood temperament may be a consideration in determining whether a child should have a "direct" treatment such as the Lidcombe Program or an "indirect" treatment based on multifactorial models (see Lectures Six and Seven). In support of their speculation, they presented preliminary results suggesting that temperament predicts outcome of an "indirect" therapy.<sup>56</sup>

The topic of clinical applicability of findings about temperament and early childhood stuttering has been debated.<sup>57</sup> The arguments presented there in favour of their clinical applicability might be overviewed as:

Stuttering is a complex, multifaceted developmental disorder with numerous research findings that highlight a wide range of individual differences and interrelations between and among multiple variables/factors .... Consequently, there is a need to obtain and integrate parent- and self-report, behavioral, and physiological data when studying, evaluating, diagnosing, and developing treatment plans for children who stutter ... Further, substantial longitudinal

evidence from other fields (e.g., psychology and child development) clearly demonstrates the interrelations between developmental challenges, parent-child attachment, emerging self-regulation processes, and socialization – with particular attention to communication on long-term (i.e., across the life span) academic, social, psychological, and vocational outcomes ... Given its heterogeneity, early attention to the child in context, as a whole, is similarly critical for understanding the onset, development, and exacerbation and/or amelioration of stuttering ... (p. 4)<sup>57</sup>

And the arguments presented there against the clinical applicability of research about temperament and early childhood stuttering might be overviewed as:<sup>57</sup>

... that constitutes the following logical fallacy: stuttering is a multifactorial disorder, therefore a multifactorial assessment and treatment approach necessarily follows. ... with the current state of evidence about temperament and early stuttering, it is not pertinent to the day to day business of providing health care to children who stutter. ... none of the data reported so far describe how pre-treatment temperament affects treatment outcome. Nor do any available data describe how clinical responses associated with a certain type of child temperament can affect treatment outcome. ... the critical clinical issue is a link between such temperament features and early stuttering development. One way to establish such a link is to discover features of temperament that distinguish stuttering from control participants prior to stuttering onset. At present, only one cohort attempted to do that, and failed, albeit with only one temperament measure and without any independent replication.<sup>[40]</sup> Emerging findings to the contrary would be a clinical game-changer. (pp. 5–6)<sup>57</sup>

Perhaps one way of summarising this clinical controversy is to present the two extreme views that have been presented by researchers and scholars. One extreme is that some children will have an unusual temperament, and some children will begin to stutter. Therefore, some children who stutter will have an unusual temperament, and how the treatment of those children needs to accommodate their temperament will be clinically obvious. The other extreme view is that temperament is involved with stuttering development in a clinically crucial way, or perhaps even a casual way, and hence temperament requires comprehensive assessment pre-treatment for satisfactory treatment to occur.

## Executive function

### *Overview of executive function*

The term *executive function* refers broadly to cognitive activity that exerts control over day-to-day activity, including thoughts, emotions, and behaviour. Executive function develops early in life. From a theoretical perspective, there is overlap between executive function and temperament. Additionally, there is empirical support for the notion that the expression of temperament may be influenced by executive function.<sup>58</sup> A review of executive function in early childhood stuttering is available.<sup>59</sup> There is another comprehensive overview of the topic and meta-analysis of research results pertinent to early childhood stuttering.<sup>60</sup> That report outlines components of executive function: *verbal short-term memory, response inhibition, attention, and cognitive flexibility*. The organisation of the present discussion of executive function and early childhood stuttering draws from this meta-analysis of children who stutter and control children. The meta-analysis involved 29 studies, 48% of which were children with early stuttering, 21% of which were school-age children, and 31% contained children from both age groups.

### *Executive function: Verbal short term memory*

Verbal short term memory can be measured with a nonword repetition task. This involves nonword sequences, which are then repeated to the examiner. Verbal short term memory can also be measured with a forward span task, where the participant is required to recall lists of numbers, letters, or words in the correct order. Nine studies showed a significant effect size for nonword repetition tasks, and seven studies showed a significant effect for forward span tasks. The authors of the meta-analysis concluded that, for verbal short-term memory, there were “robust differences” (p. 1641)<sup>60</sup> between the

child stutterers and the control children. Regardless, they tempered this conclusion by stating that those differences were subclinical, meaning that they did not require clinical attention. After the meta-analysis, another observational study of verbal short-term memory for children with early stuttering and controls reported poorer performance for the former group.<sup>61</sup>

*Executive function: Response inhibition*

*Response inhibition* is also known as *inhibitory control*. Response inhibition refers generally to “the ability to resist, subdue, or withhold one’s thoughts, behavior, and/or emotional response” (p. 1641).<sup>60</sup> With adults, Stroop tasks can be used to measure response inhibition by presenting names of colours printed in a different colour to the name and asking participants to name the colour of the printing. Several methods are available to measure dimensions of response inhibition for children of different ages. One method for young children, which involves no verbal stimulus, is the “baa-meow task.”<sup>62</sup> Children are played audio recordings of sheep and cat noises and asked to push a button that is the opposite of the animal they hear. With the meta-analysis,<sup>60</sup> five studies of parent report measures of response inhibition showed a significant effect size, and three behavioural measures showed no significant effect size.

Another method to measure response inhibition in children is the peg-tapping task. Children are asked to tap once when the experimenter taps twice, and vice versa. After the meta-analyses,<sup>60</sup> 30 child stutterers and 30 controls, aged 3–6 years, completed that task.<sup>63</sup> Significant group differences were detected, leading the authors to conclude that children who stutter “not only have difficulties with inhibition and impulsivity in the verbal domain, but also the nonverbal domain, suggesting a domain-general weakness” (p. 14). The most recent study of response inhibition with children<sup>64</sup> involved 19 Greek-speaking child stutterers and 19 controls, with a mean age of 7.6 years. No overall group differences were found for a visual task. That research group also reported a relation between inhibitory control and stuttering rate for 7-year-olds.<sup>65</sup>

When studying response inhibition with adults, researchers argued that manual response inhibition studies with adults are equivocal.<sup>66</sup> Hence, they used verbal response inhibition tasks to study 13 Cantonese-speaking adult stutterers and 14 controls. No differences were found, but they reported a significant correlation between stop-signal reaction time and stuttering severity during conversation. Although the researchers did not suggest it, such a correlation suggests that response inhibition may be a result of stuttering.

*Executive function: Attentional control*

*Attentional control* is also known as *attentional shifting*. Broadly speaking, this is the capacity to concentrate on one thing. Several aspects of this capacity can be directly measured for children using behavioural tasks and with parent report tools such as the Children’s Behavior Questionnaire.<sup>13</sup> Meta-analysis<sup>60</sup> of seven studies showed significant results for the domain of “attentional focus/persistence” (p. 1641) when measured with parent report. However, another domain of attentional control—distractibility—showed no significant effect size with parent measures for three studies. Six studies of behavioural attentional control measures were analysed, with no significant effect size. Since that meta-analysis, a report has been published<sup>67</sup> for 15 child stutterers and 18 controls, ages 8–11 years, using an experimental attention-shifting paradigm. The child stutterers had more difficulty than controls with task-shifting that required cognitive flexibility.

In this context, it is of interest that a meta-analysis involving 21 studies of attentional ability with adults who stutter<sup>68</sup> showed overall inferior performance to control participants. The author concluded that “stuttering most likely co-occurs with a problematic attentional ability only in a subgroup of PWS [people who stutter] (p. 452).”

In a review of the topic<sup>59</sup> the authors speculate about mechanisms by which “young CWS [children who stutter] would have weaknesses in executive function in the first place” (p. 311). One potential explanation is as follows:

from a resource allocation standpoint, as CWS struggle to plan or execute speech/language and/or attempt to manage their fluency breaks, they may

overutilize limited executive function resources, including aspects of attention, to compensate for fluency processes that do not come as automatically for them ... (p. 6)<sup>59</sup>

Their second explanation is based on research findings suggesting that early childhood stuttering is associated with inferior language development (see Lecture Two):

concomitant weaknesses in language processing result in limitations in executive function, which subsequently lead to deficits in other domain-specific processes. (p. 6)<sup>59</sup>

#### *Executive function: Cognitive flexibility*

*Cognitive flexibility* refers to the capacity to adapt thinking to difference circumstances. The authors of the meta-analysis<sup>60</sup> reported only two publications on this topic, hence could provide no analysis. However, the report mentioned earlier<sup>64</sup> of 19 Greek-speaking child stutterers and 19 controls found a significant group difference for a visual task.

#### *Executive function: Summary*

The term *executive function* refers broadly to cognitive activity that exerts control over day-to-day activity, including thoughts, emotions, and behaviour. There is some theoretical overlap between executive function and temperament. Mixed research findings have been published for two components of executive function: attentional control and response inhibition. This makes it difficult to form any view at present about the relevance of these constructs to early childhood stuttering. More compelling research findings have occurred for verbal short-term memory. However, in the case of those findings, their importance is difficult to assess because of their sub-clinical nature.

### **Child awareness, peer reactions, and attitude to communication**

#### *Parent-reported child awareness and negative peer reactions to early childhood stuttering*

The potential anxiety issues reported for adults who stutter can be attributed to negative social conditioning during peer interactions early in life.<sup>69,70</sup> Consequently, it is important to recognise the body of research suggesting that young children are likely to be aware of their stuttering and that it can cause them distress. This body of research contains evidence of reactions such as talking less and situation avoidance, which are interpretable as early anxiety avoidance behaviours.

An early report of children assessed by one researcher during a 6-year period documented this effect.<sup>71</sup> There were 104 children in the age range 2–4 years, and by 3 years of age around half the children were reported to show awareness of their stuttering and negative reactions to it:

Of the nine two-year-old subjects, four were said to have reacted to repetitions or other types of blockage by exclaiming, 'I can't talk,' by crying, or by looking down and blushing. In one case, seen three weeks after reported onset, the child was said to have become 'so annoyed' by his repetitions that he hit himself on the mouth and stopped talking for three days. At age three about half the children are said to have exhibited these or other evidences of reaction to stuttering blocks at one time or another. Reactions of this kind appear to be common up to about age six. At these age levels children frequently say, 'I can't talk,' 'Why can't I talk?' or 'Help me talk.' Other verbal reactions reported are 'My goodness,' 'I'm doing it again,' or 'I'll tell you later. (p. 233)<sup>71</sup>

In another early report,<sup>72</sup> five of 22 parents of children who began stuttering prior to 3 years indicated they thought their children were aware of stuttering, with four of them indicating their children were "aware and bothered" (p. 176) by it.

Later reports have been consistent with these findings. One showed that 57% of 1,122 parents of 2-year-olds said that their children showed signs of awareness of stuttering, and the figure had increased to 90% for 7-year-olds.<sup>73</sup> Around two-thirds of children who had been stuttering for less than 1 month reportedly showed awareness of it. Using a Japanese version of the survey in the first study, that result was replicated with 57 Japanese children of ages 3–7 years.<sup>74</sup> Seventy percent of parents reported

child awareness of stuttering at 4 years, and 90% at 7 years. Another report of 77 parents of children, mean age 53 months (range 34–73 months),<sup>75</sup> showed that 90% of them reported some kind of negative impact from stuttering. The most common reported reactions were “frustration associated with their stuttering, withdrawal, reduced or changed verbal output, making comments about their inability to talk” (p. 407).<sup>75</sup> Twenty-five per cent of the parents linked stuttering with talking less, and 43% said stuttering negatively affected the children’s mood. Twenty-seven per cent reported that peers teased their children about their stuttering, which is a higher rate of teasing than the usual 6–22% range reported for children who do not stutter.<sup>76,77</sup> Thirty-eight Norwegian parents of 2–5-year-olds used an adaptation of the OASES to report the impact of stuttering on their children.<sup>78</sup> Results indicated that most parents thought that stuttering adversely affected their children.

Parent-reported frustration with stuttering is consistent with that experienced by adults, as noted at several places during these lectures. A direct connection between early stuttering and frustration was measured in a study of 13 children<sup>79</sup> who stuttered and 12 controls, mean ages 6 years and 5 months. An experimental frustration task revealed that the children who stuttered showed less frustration tolerance than the controls.

#### *Attitude to Communication*

The KiddyCat<sup>80</sup> is designed as a measure of attitude to communication. Children are asked 12 *yes/no* questions, such as “is it hard for you to say your name,” “do your words come out easily,” “do mom and dad like how you talk,” and “do people like how you talk?” (p. 229).<sup>80</sup> The KiddyCat might be interpreted as having some relation to social anxiety (see Lecture Twelve).

Three studies from the United States have shown KiddyCat score differences between children with early stuttering and controls: with 52 stuttering and 62 control children,<sup>81</sup> 45 stuttering and 63 control children,<sup>82</sup> and with 46 stuttering and 66 control children.<sup>83</sup> The same result has been reported for 58 stuttering and 70 control Polish children,<sup>84</sup> for 49 stuttering and 74 control Slovenian children,<sup>85</sup> and 55 stuttering and 53 control Turkish children.<sup>86</sup> The KiddyCat was used in a study mentioned previously<sup>21</sup> with small participant numbers, and it showed no differences between stuttering and nonstuttering children. A report of 59 children with a mean age of 4.8 years (range 2–6) years<sup>87</sup> indicated that neither stuttering severity nor time since reported stuttering onset predicted KiddyCat scores. A report of 296 pre-schoolers with a mean age of 47.9 months<sup>88</sup> found that KiddyCat scores were related to age, but essentially not related to behavioural features of stuttering.

#### *Evidence of negative peer responses*

There is direct video evidence of negative peer reactions to stuttering in pre-school playgrounds.<sup>89</sup> Four children with early stuttering were video recorded for four 20-minute playground periods. During these 80-minute samples of conversation, negative peer responses to stuttering were reported for three of the four children. The percentages of negative peer responses to stuttered utterances that had communicative intent for the three children were 2.8%, 12.5%, and 28.6%. For the latter child, more than a quarter of stuttered utterances with communicative intent received negative peer responses. Those peer responses included interrupting, mocking, walking away, and ignoring what the child was saying. One child was even assaulted because his stuttering prevented him from resolving a conflict about a toy.

Those results are alarming because, if they in any way reflect what generally happens to young children who stutter, there is good reason to believe that they may be exposed to the kind of negative social conditioning that could be the origins of anxiety later in life.

#### *Peer awareness*

Indirect evidence that children with early stuttering may receive negative social conditioning comes from an ingenious research paradigm involving stuttering and nonstuttering puppets.<sup>90</sup> On three occasions over a 2-year period, 20 stuttering and 20 nonstuttering pre-school children were asked to “point to the puppet that talks the way you do” (pp. 233–234). Results showed that the children were generally able to identify with the stuttering and control puppets, and their reliability for so doing

increased during the period of study.

A preliminary report has begun for a method to explore pre-schoolers' attitudes to stuttering using avatars.<sup>91</sup> This method adapts the well-known *Public Opinion Survey on Human Attributes—Stuttering* (POSHA-S) that has featured in much of the research about stuttering stereotypes discussed during Lecture One. Preliminary results with the POSHA-S/Child for 51 children 3–7 years old indicated the potential for negative attitudes toward stuttering at that time of life. A subsequent report<sup>92</sup> verified this finding. Children 4–10 years old generally had more negative attitudes than adults but showed systematic improvement during that period. That improvement trend was replicated with children and parents from Bosnia and Herzegovina.<sup>93</sup> The POSHA-S/Child was used to assess attitudes to stuttering of 37 pre-schoolers before and after an intervention designed to improve it.<sup>94</sup> The intervention comprised two 30-minute classroom lessons with puppet videos, group discussion, and schoolbook activities. Results showed a post-intervention improvement of attitudes. The result was replicated with Polish children.<sup>95</sup>

### A clinical trial of resilience training for stuttering with pre-school children

#### *Background*

The concept of psychological resilience has been defined as “the process and outcome of successfully adapting to difficult or challenging life experiences, especially through mental, emotional, and behavioral flexibility and adjustment to external and internal demand.”<sup>96</sup> In other words, “bouncing back” after adversity. Resilience develops during childhood, and there are data to suggest that, with 5–18-year-olds, resilience is related to the impact of stuttering measured with the OASES.<sup>97</sup>

#### *Background*

The authors of this clinical trial<sup>98</sup> based their design on material discussed earlier in this lecture. They cited “strong evidence of the significant differences in self-regulation, emotional reactivity, and resilience” (p. 71) between pre-schoolers who stutter and those who do not. They designed their trial for many purposes, including to determine whether speech treatment improves after intervention and the effects of resilience training on pre-schoolers who stutter and their parents. However, a key issue explored with the trial was raised earlier by those involved with research about early childhood stuttering and temperament: whether a resilience component added to speech treatment for children with early stuttering improves speech outcomes.

#### *Design*

The randomised controlled trial was used to compare a group of pre-schoolers who received speech treatment for stuttering compared with a group who received that speech treatment and “an additional resilience component” (p 71).<sup>98</sup> Participants were 28 child-parent pairs with children being a mean age of 4.4 years. Assessments occurred pre-treatment and immediately after 12 weeks of treatment.

#### *Outcomes*

Four standard outcome measures were used, but none were specified as the primary outcome. Percentage of syllables stuttered was obtained from a within-clinic sample of 350–450 syllables during a play task. The Parenting and Family Adjustment Scales<sup>99</sup> measure various domains of family functioning and parenting, and the parenting practices domain was used. The Strengths and Difficulties Questionnaire<sup>3</sup> measures emotional and behavioural problems. Finally, the authors used an adapted version of the effortful control subscale of the Children's Behavior Questionnaire.<sup>100</sup>

#### *The treatments*

The speech treatment was “a combination of direct and indirect stuttering therapy” including “response contingent principles,” “‘Demands and Capacities Model’ treatment” (p. 74.)<sup>98</sup> (see Lecture Three), and Palin Parent-Child Interaction therapy (see Lecture Six). The additional component in the

experimental treatment arm was the Curtin Early Childhood Stuttering Resilience Program.<sup>†</sup>

### *Results*

Overall, the pre-treatment mean %SS was 10.5 and post-treatment was 2.1. There was no evidence that the resilience treatment improved these outcomes. There was no evidence of any improvement post-treatment for the temperament dimension of effortful control. There was evidence of reduced behavioural and emotional problems in both groups, but no evidence that the child resilience training added to that effect. For both groups, there was evidence of improved parenting practices at post-treatment. However, there was evidence that the experimental group that received parent training had more improvement than the group that did not.

### *Limitations*

The authors conclude that “this study provides evidence for the overall effectiveness of early intervention in stuttering therapy” (p. 79). Yet the extent of the contribution is limited by the evaluation of the treatment, which was based on 350–450 syllables of within-clinic speech measured immediately post-treatment without a follow-up period. According to some criteria, the report would not be regarded as a clinical trial (see Lecture Five). The authors also conclude that the experimental treatment was “successful in positively shifting parenting practices and developing improved self-regulation and resilience” (p. 79). It might be arguable that a more important contribution of the trial is the finding that such changes had no impact on speech treatment outcomes. It is also difficult to interpret the results of the study because there was no indication of whether pre-treatment scores for the Parenting and Family Adjustment Scales, the Strengths and Difficulties Questionnaire, and the Children’s Behavior Questionnaire were within or beyond the typical clinical range.

### **Conclusions: The origins of social anxiety with stuttering during the early years**

It seems clear that children are likely to be aware of stuttering shortly after onset and that it may well cause them distress. Additionally, early childhood stuttering may be associated with negative peer social conditioning, which is potentially associated with anxiety development later in life. Direct test evidence of psychological problems with pre-school children who stutter so far contains conflicting reports. One of four small studies reported a difference between stuttering pre-schoolers and controls. The ELVS cohort showed no differences from controls for the Strengths and Difficulties Questionnaire during the pre-school years, but the Millennium cohort and the United States National Health Interview Survey showed that there were differences. Such findings so early during the developmental course of stuttering cannot be disregarded.

The ELVS cohort, although yet to be replicated and with limited methods to measure temperament, shows that children who begin to stutter show no signs of temperament markers of anxiety prior to or during early stuttering development. On balance, there is no consistent pattern of evidence that pre-school children who begin to stutter have temperamental markers of anxiety. Hence, findings of an association between early childhood stuttering and unusual temperament are easily interpreted as an epiphenomenon reflecting the effects of stuttering, or simply as a benign co-occurrence with it. As an example of the latter possibility, it would not be surprising if children with an anxiety-prone temperament became more anxious in response to early childhood stuttering than children who did not have such a temperament.

On balance, it seems that anxiety, autonomic nervous system involvement, temperament, and executive function are associated somehow with early childhood stuttering. Yet, to date, that body of literature has yet to yield an overarching and generally endorsed explanation of this material that is clinically useful. One clinical trial has included that material but not provided definitive results. On

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<sup>†</sup> The authors note a study limitation that this intervention has not been standardised and there is no evidence of its efficacy. It has not been reported previously and no references to it, or to a treatment manual, are cited in the report. The paper incorporates a similarly unreported and unreferenced pilot assessment connected to the intervention: the Curtin Early Childhood Stuttering Resilience Scale. Again, the authors acknowledge a study limitation that the scale has not been assessed for reliability and validity. Consequently, results from the scale are not discussed here.



the face of it, there are two broad possibilities that might emerge during coming years: Either the findings are an epiphenomenon, or they reflect something about the nature and cause of stuttering. At present, it seems compelling to conclude the former, and that that anxiety-related mental health issues are a consequence of early childhood stuttering rather than being involved in its causality.

A report is available for a 2022 symposium that dealt with the clinical application of research findings about mental health and early stuttering.<sup>101</sup> Master clinicians presented their responses in a clinical situation where parents of a 3-year-old who stuttered asked about associated early signs of mental health issues. Then, a discussion occurred about the matter between the master clinicians, a distinguished scholar, and conference delegates.

The scholar's views, which were accepted by the discussants, were consistent with the research overviewed in this lecture. There is no evidence of "anxiety proneness, temperament, or personality" (p.7) existing before stuttering onset, hence there is no reason to believe that they are part of a causal model of stuttering. Regardless, "there is some connection between early stuttering and temperament" (p. 7). Two clinicians took a pragmatic view of this matter; it is not surprising that early stuttering might be distressing for children and parents, so empathy and support should be provided as needed. However, a third clinician expressed a perspective that:

Working with parents to address their worries and to support the child, encouraging openness in the family, and fostering a positive attitude to stuttering, will be key in any treatment trying to avoid or minimize long-term psychological impact (p. 7)<sup>101</sup>

The issue was raised that if one of the early interventions discussed during Lectures Six and Seven is effective, that may prevent further development of psychological problems with early stuttering. Regardless, one clinician made the important point that every child who stutters will experience the condition for a period, during which negative psychological consequences might occur.

## **THE ORIGINS OF SOCIAL ANXIETY WITH STUTTERING: THE SCHOOL-AGE YEARS AND ADOLESCENCE**

### **Anxiety and anxiety-related measures**

#### *General Anxiety Scale for Children*

The 1,000-family study<sup>102</sup> (see Lecture Two) presented extensive early data about anxiety with a group of 80 school-age child stutterers and 80 controls ages 9–11 years, with mean age 10.5 years. The children received an extensive psychiatric evaluation, including the General Anxiety Scale for Children.<sup>103</sup> A limitation of those data, however, was that speech-language pathologists rather than psychologists or psychiatrists collected them.

#### *State-Trait Anxiety Inventory*

It was 30 years until further data about this matter emerged.<sup>104</sup> In the context of a clinical trial, the State-Trait Anxiety Inventory for Children<sup>105</sup> showed no significant pre-treatment differences between 77 stuttering and 20 control children, ages 9–14 years, with mean age 10.9 years. Neither group was unusual according to test norms. A subsequent report<sup>106</sup> replicated this finding with the same stuttering participants and an enlarged group of 106 control children 9–14 years old with mean age 11 years.

Another report<sup>107</sup> used the State and Trait Anxiety Inventory for Children with three groups who were slightly older: 18 who stuttered and were seeking treatment (10–16 years, mean 12.6 years), 17 who once stuttered but recovered (10–16 years, mean 12.7 years), and 19 controls (10–15 years, mean 12.9 years). There was no difference between the groups for trait anxiety, but the stuttering group scored higher for state anxiety in three of four hypothetical situations that were used in the test.

Another report<sup>108</sup> used the State Trait Anxiety Inventory<sup>109</sup> (for adults) with an older group: 19 stuttering adolescents who were seeking treatment and 18 controls between 11–18 years, with a mean age of 14.4 years. Results showed significantly higher state and trait anxiety scores for the stuttering group.

*Fear of Negative Evaluation Scale*

The report just mentioned<sup>108</sup> used the long version of the Fear of Negative Evaluation scale<sup>110</sup> with adolescents (mean age 14.4 years) and reported significantly higher scores for the stuttering group. However, a caveat to this finding is that the Fear of Negative Evaluation scale was developed for adults, not adolescents.

*Revised Children's Anxiety and Depression Scale*

A report of 132 child stutterers, ages 9–14 years,<sup>42</sup> who presented at a speech clinic, were given the Revised Children's Anxiety and Depression Scale.<sup>43</sup> The scale has a child- and a parent-reported version, and measures anxiety and depression symptoms in 8- to 18-year-olds. It has five subscales related to anxiety. All the children's scores were below clinical thresholds.

*Revised Children's Manifest Anxiety Scale*

Revised Children's Manifest Anxiety Scale<sup>111</sup> data were reported with 18 stuttering and 18 control children in the age range 11–12 years.<sup>112</sup> Although some significant results were reported for the total anxiety and subscale scores, these results could not be interpreted as clinically significant because the child stutterers were not beyond the typical range of scores. Another report<sup>113</sup> using the Revised Children's Manifest Anxiety Scale involved 36 stuttering adolescents who had received treatment, and 36 controls aged 12–18 years, with a mean age of 14.3 years. As was the case with school-age children, the stuttering adolescents scored significantly higher. However, both groups were in the typical range for anxiety.

A report<sup>114</sup> of 23 stuttering school-age boys and girls, ages 6–11 years, and 50 adolescent boys and girls, ages 13–18 years, produced consistent results. The total anxiety score and subscale scores were within typical limits. However, the Revised Children's Manifest Anxiety Scale contains a lie scale, which is designed to detect deceptively positive responses where respondents present themselves in a favourable light. There was evidence that the boys had high lie scale scores, suggesting they may have been concealing their true levels of anxiety. The researchers speculated that this might explain the many equivocal results about anxiety levels in these age groups.

*Personal Report of Communication Apprehension*

The Personal Report of Communication Apprehension<sup>115</sup> deals with fear of speaking, with subscales for public speaking, meetings, groups, and conversations. A study<sup>116</sup> recruited 39 stuttering adolescents from speech clinics along with 39 controls, ages 13–18 years with a mean of 14.6 years. The adolescents with stuttering showed significantly higher apprehension scores than controls. This result was replicated with a study<sup>117</sup> of 36 adolescents seeking stuttering treatment, ages 11–18 years, mean age 14.2 years. There were no controls but results for the adolescents were consistent with the first study.<sup>116</sup>

*Strengths and Difficulties Questionnaire*

The Strengths and Difficulties Questionnaire mentioned earlier<sup>3</sup> was used in a study<sup>118</sup> with 10 boys who stuttered and 10 controls, ages, 8–14 years. The authors reported a significant difference between the groups. The previously mentioned report from the Millenium Cohort<sup>7</sup> used the Strengths and Difficulties Questionnaire to show that 11-year-olds (N=194) reported to be stuttering by their parents differed from matched controls. The questionnaire was used in the United States National Health Interview Survey report mentioned earlier,<sup>6</sup> and significant differences were found for some survey domains with children ages 6–10 years and 11–17 years. Another report, with 35 boys who stuttered and 35 controls, ages 14–17,<sup>119</sup> found a significant difference for the survey domain dealing with peer relationships.

*Child Behaviour Checklist*

A study of Turkish child stutterers and controls<sup>25</sup> included children in the ages 7–11 years and 12–18 years, with 15 stuttering children and 15 controls in both age groups. The Child Behaviour Checklist<sup>11</sup> was used and significant differences were found between the groups for nearly all eight scales of the

test, notably “Anxious/Depressed.”

#### *The Perseverative Thinking Questionnaire*

The authors of the Perseverative Thinking Questionnaire<sup>120</sup> define perseverative thinking as repetitive, intrusive and “difficult to disengage from” (p. 226), and report that it is related to anxiety and depression. A similar construct is described as “rumination” in the Clark and Wells model of social phobia, as described in Lecture Ten. A report of 313 adult stutterers<sup>121</sup> linked repetitive negative thinking with OASES-A scores and certain aspects of temperament. Subsequently, those researchers explored repetitive negative thinking with 99 children and adolescents in the age range 9–18 years.<sup>122</sup> All participants except one had received treatment for stuttering. A version of the Perseverative Thinking Questionnaire for 9–15-year-olds was used,<sup>123</sup> along with the OASES-S and OASES-T, and a screening tool for anxiety-related disorders in childhood.<sup>124</sup> Additionally, participants were asked to respond from “never, rarely, sometimes, often, always” to the statement “my goal when speaking is to stutter openly and not do anything to try to hide it” (p. 5).

Results were that 22% of participants screened positive for generalized anxiety disorder and 32% screened positive for social anxiety disorder. Associations were reported between repetitive negative thinking and OASES scores. Associations were reported between screening for generalized anxiety disorder, social anxiety disorder, OASES scores, and repetitive negative thinking. There was a small but statistically significant association between repetitive negative thinking and responses to the statement “my goal when speaking is to stutter openly and not do anything to try to hide it” (an odds ratio of .96).

#### *Speech Situation Checklist-Emotional Reaction*

A report<sup>125</sup> presented data from the Speech Situation Checklist-Emotional Reaction<sup>126</sup> for 100 child stutterers and 275 controls, aged between 7–14 years. The test assesses anxiety associated with various speaking situations and was translated into Kannada for the study. Kannada is spoken by around 70 million people in the south of the Indian subcontinent. The child stutterers had double the scores attained by the control children. Additionally, there was a developmental trend, with the older child stutterers in the cohort having much higher scores than the younger children.

### **Multiple Anxiety Assessments**

A report<sup>127</sup> presented a range of assessments for 37 adolescents who were seeking treatment for stuttering, ages 12–17 years and mean age 14.2 years. They were given a computerised, self-administered version of the Diagnostic Interview Schedule for Children.<sup>128</sup> Fourteen of them (38%) met diagnostic criteria for at least one DSM-IV<sup>129</sup> mental health disorder,<sup>†</sup> which is around twice the anticipated rate for adolescents, placing it in the 17–21% range.<sup>130,131,132</sup> Ten of these 14 diagnoses were a mental health disorder involving anxiety: social anxiety disorder, agoraphobia, obsessive-compulsive disorder, separation anxiety disorder, and specific phobia.

For three psychological test scores, the stuttering participant scores were in the typical range: Revised Children’s Manifest Anxiety Scale,<sup>111</sup> Child Behavior Checklist,<sup>11</sup> and the Children’s Depression Inventory.<sup>133</sup> However, there was a consistent trend for the older adolescents (15–17 years) to have more severe scores than the younger adolescents (12–14 years). A report described 65 adolescents who stuttered, ages 14–18 years, and who presented to a child and adolescent psychiatry clinic.<sup>134</sup> There were significant correlations between stuttering severity, depression, and social anxiety. For scores on the Beck Depression Inventory,<sup>135</sup> 20% of participants showed moderate depression, and 28% showed severe depression. However, this report needs to be interpreted cautiously because of the biased sampling from a psychiatry clinic.

A study of 75 stuttering 7–12-year-olds and 150 matched nonstuttering controls<sup>136</sup> included the Youth Online Diagnostic Assessment.<sup>137</sup> Results indicated that, compared with controls, the stuttering group

<sup>†</sup> The DSM-IV is the previous edition of the DSM-5, which was mentioned earlier.

had four-fold increased odds for prevalence of any anxiety disorder. For social anxiety disorder there was six-fold increased odds, with girls much more at risk than boys and with 24% of the child stutterers diagnosed with the condition.

The Spence Children's Anxiety Scale Child Report<sup>138</sup> and Parent Report<sup>139</sup> showed scores within the typical range, but significantly higher values for the stuttering group. Mean scores for the Strengths and Difficulties Questionnaire Parent Report<sup>140</sup> were within typical limits, however the mean Total Difficulties score and Internalising and Externalising scores were significantly higher for the child stutterers.

The largest cohort to date<sup>141</sup> involved 102 adolescents, ages 11–17 years, who were seeking treatment for stuttering. Psychological test scores were reported for the Revised Children's Manifest Anxiety Scale,<sup>111</sup> the Children's Depression Inventory,<sup>133</sup> the Youth Self Report and Child Behavior Checklist,<sup>11</sup> and the Assessment of the Child's Experience of Stuttering, which is an earlier version of the OASES-S. Stuttering severity measures were reported and showed that the relationship between them and psychological measures was not straightforward. Scores for depression and anxiety were within typical limits, but higher self-reported stuttering severity was associated with higher anxiety and internalising (emotional) problems. The boys showed externalising problems (rule-breaking and aggression) in the clinical range.

A report<sup>142</sup> from the ELVS cohort (see Lecture Two) when the children were 11 years old compared those who were stuttering with those who were not, using the Spence Children's Anxiety Scale Parent and Self Report,<sup>138,139</sup> the Strengths and Difficulties Questionnaire,<sup>3</sup> and the School-Aged Temperament Inventory.<sup>143</sup> In contrast to the study of other Australian children reported around the same time,<sup>141</sup> no differences were found. A likely cause of that discrepancy is that one study involved children from a clinic and the ELVS report did not.

### Physiological evidence

A study<sup>144</sup> of nine child stutterers, ages 6–11 years (mean age 9.3 years), without a control group, compared salivary cortisol with normative data. Measures were made four times per day for three consecutive days. No evidence of atypical levels was found; however, the authors noted that future research might take account of the many potentially confounding methodological issues with making such measures during childhood.

Anticipation of stuttering is common among those affected and is linked to anxiety (see Lecture One). It is possible—even likely—that unusual eye gaze patterns during reading are a physiological marker showing anticipation of certain words, and there is evidence that adults who stutter show such unusual eye gaze patterns during reading (see Lecture One). Three reports have shown that school-age children have such eye gaze patterns that are potentially consistent with anticipation of difficulty with certain words.<sup>145,146,147</sup>

### Communication attitude

Negative communication attitude has been documented not only for pre-schoolers as outlined previously, but also for older children and adolescents. A detailed review of the topic is available.<sup>148</sup> The Communication Attitude Test, often referred to as CAT, is the original scale developed for primary school-age children from which the KiddyCAT—described earlier—was derived. As with the KiddyCat, the CAT may have some relation to social anxiety (see Lecture Twelve).

There is a substantial body of research showing that children from several cultures and languages who stutter have higher scores than controls for this test. The Communication Attitude Test was used to assess 70 stuttering and 271 control 7–14 year old Belgian children.<sup>149</sup> From age 7 years the child stutterers had more negative attitudes to communication than peers. Additionally, their communication attitude worsened with time, whilst it became healthier with the controls. Other reports with 143<sup>150</sup> and 110<sup>151</sup> Belgian school-age children have replicated these findings.

These Communication Attitude Test differences have been replicated with Italian 7–14-year-olds<sup>152</sup>

and Croatian 7–13-year-olds.<sup>153</sup> Eighty stuttering and 80 control Japanese school-age children, ages 5–12, also showed differences,<sup>154</sup> and there was evidence that the scores of the stuttering group continued to worsen across the ages studied, while the control group seemed to stabilise. Similar results have been found with Swedish<sup>155</sup> and Slovenian<sup>156</sup> children.

### Perceived communication competence

For those affected by stuttering during adolescence, the dimension of perceived communication competence might also intuitively suggest an indirect relation to social anxiety. There have been several reports using the Self Perceived Communication Competence scales.<sup>157</sup> The study that reported about communication apprehension<sup>116</sup> also reported data from the Self Perceived Communication Competence scales for the 39 stuttering adolescents from speech clinics and 39 controls (ages 13–18 years with a mean of 14.6 years). The stuttering group had significantly poorer perceived communication competence than the control group. The same research team replicated these significant results<sup>158</sup> with adolescents 13–18 years old: a group of 53 receiving stuttering treatment (mean age 15.2 years) and 53 controls (mean age 14.8 years). The previously mentioned study of 36 adolescents<sup>117</sup> found similar results with the Self Perceived Communication Competence scales. Consistent results were found with interviews comparing stuttering and control children ages 5–10 years.<sup>159</sup>

### General psychological distress

A birth cohort study<sup>160</sup> identified 217 adolescents who were stuttering at 16 years, according to their parents, 137 of whom completed the Rutter Malaise Inventory.<sup>161</sup> This is not a specific measure of social anxiety but “a 24-item self-completion scale which measures emotional distress such as depression and anxiety and related somatic symptoms such as headaches and tiredness” (p. 459).<sup>161</sup> The clinical history of the participants was not reported. Results showed that the adolescents with stuttering were more likely than the controls from the cohort to experience psychological distress, but not at levels that would put them at risk for clinically significant mental health disorders. However, one study has reported positive levels of self-esteem for 48 adolescents who stuttered,<sup>162</sup> and another<sup>163</sup> has reported the opposite effect for 54 participants and controls.

### Peer awareness

The potential anxiety issues reported for adults who stutter can be attributed to negative social conditioning during peer interactions early in life.<sup>69,70</sup> The earliest report that school-age children are aware of stuttering in peers was in 1958 with 120 children 5–8 years old.<sup>164</sup> They were presented with recordings of a story with and without stuttering, and the older children showed a preference for the latter and sometimes used the term “stuttering.” An independent group replicated these results with a similar method two decades later.<sup>165</sup> In this study, 30 nonstuttering children, mean age 6.7 years, preferred to hear a story that was told without stuttering rather than a story told with stuttering. A second group of nonstuttering children, mean age 8.9 years, not only preferred the nonstuttered story, but volunteered the label “stuttering” when describing the other story.

The puppet research method described earlier for pre-schoolers was used with 79 nonstuttering children ages 3–7,<sup>166</sup> and reported consistent results. The children were able to identify with the nonstuttering puppet, again with that capacity increasing with age. It was telling that, from 4 years of age, the children began to offer negative evaluations of the stuttering puppet.

A study with 75 children, mean age 9 years 10 months, used the semantic differential bi-polar adjective pair method.<sup>167</sup> Half the children watched a video of an adult stuttering, and the other half watched the same adult not stuttering. For 12 personality attributes, the children assigned significantly more negative scores for the video with stuttering. A study of 64 school children aged 10–14 years, mean 12.7 years, involved videos of a peer speaking at four different stuttering severities.<sup>168</sup> The children rated the videos for a range of attributes dealing with “themes of peer friendship, listener comfort, and allowing a peer who stutters to take a speaking role in a group project” (p. 208).<sup>168</sup> There was a significant relationship between stuttering severity and the negativity of the peer responses. A

similar study with 88 children ages 8–12 years reported the same relation between negative perceptions and stuttering severity.<sup>169</sup> A study of 62 Italian school child stutterers and 474 controls, with mean age 11 years, reported that the child stutterers were more rejected by and less popular with peers.<sup>170</sup> However, a report<sup>171</sup> of a small sample 22 stuttering school children, with a mean age of 14 years, in Flanders (Belgium) showed no evidence of peer rejection.

### Bullying

Generally, being bullied during the school years is strongly associated with anxiety later in life,<sup>172</sup> and one report shows this effect concurrently for a control group and a group of adults with stuttering.<sup>173</sup> Hence the association of bullying with stuttering school-age children is of interest in the present context. One report<sup>112</sup> showed stuttering school-age children to have a 63% risk of being bullied compared to 22% for controls. Another report<sup>174</sup> of 28 child stutterers, ages 7–15 years, found that 59% of them reported being bullied, and 38% reported it to have occurred on most days or every day. And another report<sup>117</sup> of 36 adolescents indicated that 63% reported being bullied less than once per week, but 37% reported it occurring at least once per week. With a study of 53 stuttering adolescents and 53 controls,<sup>158</sup> aged 13–18 years, with a mean age of 15.2 years, more adolescents reported being bullied: 43% compared to 11% for controls. The study discussed earlier,<sup>136</sup> with 75 stuttering and 150 control children, indicated higher scores for the child stutterers on the Culture Bullying scale of the Personal Experiences Checklist Child Report.<sup>175</sup> The following two items were responsible for the significant result: “other kids make fun of my language” and “other kids tease me about my voice.” A report of 54 stuttering adolescents and 54 controls<sup>163</sup> indicated significantly more bullying for the former group using The Bully-Victimization Scale.<sup>176</sup>

Another report<sup>177</sup> concerned 403 nonstuttering children, mean age 11 years 9 months, who each had a stuttering classmate. The children were asked to categorise their stuttering peers. One of the categories was “bully victim,” and 38% of child stutterers were placed in that category compared with only 11% of nonstuttering children. Consistent with that finding, classmates thought stuttering peers to be less popular, without leadership potential, and more likely to be rejected than others. A study of 97 school children, aged 8–13 years<sup>178</sup> with a mean age of 10 years, used the Peer Attitudes Toward Children Who Stutter scale.<sup>179</sup> The study reported that children who had contact with a stuttering child had significantly more negative attitudes to child stutterers. The same research group replicated that finding with 760 children ages 6–12 years.<sup>180</sup> The presence of bullying in classrooms has been confirmed with a report by elementary to high school United States teachers.<sup>181</sup>

Retrospective reports of bullying by adults have produced results consistent with the above findings. In one report,<sup>182</sup> pertaining mostly to the school years, 83% of 276 stuttering adults reported being bullied at school, with 18% reporting it occurred every day and 41% reporting a few times per week. Almost all respondents reported negative short-term effects of being bullied, and 46% reported long-term effects. These results were replicated with a survey of 324 adult respondents,<sup>183</sup> 82% of whom reporting being bullied at least once per week. Responses suggested that 84% of respondents had difficulty establishing friendships later in life because of the bullying. One report of 332 adults surveyed<sup>184</sup> reported that 56% said they were “affected a lot” by stuttering during the school years. There is evidence that such lasting effects are associated with cyberbullying.<sup>185</sup>

### Impact of stuttering

These findings about bullying and mental health with stuttering school-age children are consistent with a report using the OASES impact measure for 50 stuttering 8–11-year-olds, and an adapted version for controls.<sup>186</sup> The adapted OASES version replaced the word “stuttering” with “speaking ability.” The child stutterers had significantly lower impact scores than peers. This was reflected in overall concern about their speech, increased behavioural and cognitive responses to their stuttering, and compromises to their communication in daily situations. A study of OASES scores for 162 participants<sup>187</sup> involved children, adolescents, and adults. All scores were mostly in the moderate impact range (2.25–2.99). Statistically significant trends were found, including higher scores for adolescents than children and adults. Female participants were impacted more than males, and

adolescent girls were impacted by their stuttering more than adolescent boys. The mean OASES score for girls was 2.72, and for boys was 2.37.

### Conclusions: Anxiety during the school-age years and adolescence

In contrast to pre-school children who stutter, there is much more evidence that the anxiety-related mental health issues that affect adults who stutter begin during the school-age and adolescent years. There are signs that such problems develop and worsen during this period, with findings of problem anxiety measures more typical of older participants in studies. Two reports<sup>127,136</sup> have found evidence during the primary school years and adolescence of the diagnosable anxiety-related mental health disorders that trouble adults. The latter of these reports found evidence that 24% of school-age children presenting at speech clinics for stuttering treatment were diagnosed with social anxiety disorder. The earlier of these reports contained evidence of worsening anxiety test scores during adolescence. Evidence of bullying during the school years, and negative classroom experiences, are consistent with these findings. The most prominent anxiety disorder with adults who stutter—social anxiety disorder—is typically diagnosed during early adolescence,<sup>188,189</sup> with median onset at 13 years.<sup>190</sup> So it is not surprising that it is present for many school-age children who stutter, warranting referral to a clinical psychologist.

These findings about the early psychological effects of stuttering are consistent with a body of evidence that children who have speech and language disorders are generally at risk of developing mental health problems, many of them involving anxiety.<sup>191</sup> One of those reports<sup>192</sup> was of 258 five-year-olds who were diagnosed with a speech or language disorder (only five were diagnosed with stuttering). Controls had a 21% rate of psychiatric disorder, and the language-impaired group had twice that rate at 40%. At a 14-year follow-up the rate of psychiatric disorder had not changed. The authors concluded that “young adults with a history of early childhood language impairment have one of the highest rates of psychiatric disorder in the community” (p. 80).<sup>192</sup> That could certainly be said of stuttering.

## STUTTERING, MENTAL HEALTH, AND THE TIMING OF EARLY INTERVENTION: A POSITION STATEMENT

Early intervention is by far the best clinical option for stuttering, as outlined during Lectures Six and Seven. Considering epidemiological data and evidence of the potential quality of life impairment from persistent stuttering, and the mental health evidence presented during this lecture, the following position statement about the timing of early intervention seems justified.

Stuttering typically starts during the pre-school years and is associated with significant risk of mental health problems later in life, particularly social anxiety disorder. Such problems have been reported from 7 years of age and are associated with long-term impairment of educational and occupational attainment. The origins of those mental health problems have been reported during the pre-school years for children who stutter: negative peer reactions, teasing, stigmatisation, social distress, and signs of emotional and behavioural problems. Although three-quarters of children may eventually recover naturally from stuttering sometime later in life, recovery rate during the first 18 months is estimated to be only 6–8%, as discussed during Lecture Two. However, it is not possible to predict whether an individual child will recover naturally. Consequently, after diagnosis, stuttering should be treated with an appropriate evidence-based treatment as soon as possible.

That position does not appear to be universally endorsed. A report of a 2019 symposium about stuttering treatment,<sup>193</sup> with delegates from 29 countries, raised the prospect of “active monitoring” after stuttering onset, rather than beginning an evidence-based treatment immediately. The conference convenors concluded as follows:

However, one issue remained unresolved among us: whether to intervene immediately after onset or to delay intervention. And in the event that a decision is made to delay intervention, it seems that some clarity is needed about what “active monitoring” means and how it fits into that management

plan. Is it pre-treatment counselling, is it a treatment intended to reduce stuttering, or is it a procedure to determine whether natural recovery occurs? (p. 9)<sup>193</sup>

Another report<sup>194</sup> that included 126 Norwegian speech-language pathologists, appeared to provide a somewhat different perspective. When surveyed about this matter with pre-school children, two-thirds of the speech-language pathologists “reported that they never recommend a ‘wait and see’ approach when they are initially contacted by a parent reporting that their child is stuttering” (p. 929).

A report of a 2022 symposium<sup>195</sup> resumed the topic of natural recovery and early intervention. A clinical situation was role-played where parents of a 3-year-old asked a clinician whether recovery from early stuttering without treatment can be predicted. There was general agreement between scholars and clinicians that such recovery cannot be predicted. Regardless, none of the clinicians responded to that clinical situation by recommending immediate intervention for the purpose of expedient recovery. Issues underlying that decision were (1) doubts about the efficacy of early intervention, (2) weighing up the benefits of direct early intervention against the practical imposition of those methods on families, and (3) whether the stuttering appeared to be affecting the child and family negatively.

## SUMMARY

There is strong evidence that social anxiety issues with adults who stutter begin early in life. There is direct and indirect evidence that such issues begin during the pre-school years. Temperament is involved with early childhood stuttering, but it is not clear at present whether that involvement is theoretically and clinically important, or whether it is an epiphenomenon. Primary school age and adolescent clients seeking stuttering treatment are more likely than younger clients to experience clinically significant anxiety. Peers notice their stuttering and they are bullied more often than peers. Speech-language pathologists need to take account of these findings when making decisions about the timing of early intervention. There is a strong case to argue that, after diagnosis, early childhood stuttering should be treated with an appropriate evidence-based method as soon as possible.



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## LECTURE TWELVE: TREATMENT OF SOCIAL ANXIETY AND STUTTERING<sup>†</sup>

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### SPEECH-LANGUAGE PATHOLOGISTS AND ANXIETY TREATMENT

As noted during the previous lectures, some clients presenting at speech clinics with stuttering may have clinically significant anxiety that requires intervention. This has prompted statements that any such client with significant anxiety, regardless of whether it amounts to a diagnosable psychological problem, requires clinical management.<sup>1,2</sup>

That said, not all clinicians will have the necessary training or experience to manage the social anxiety of stuttering clients. Management of anxiety is fundamentally in the professional domain of clinical psychologists and psychiatrists. Speech-language pathology professional preparation programs around the world vary in the extent to which they incorporate anxiety management training. However, alone, they are not a qualification to diagnose and manage anxiety disorders.

The authors of a tutorial about anxiety management procedures for stuttering clients<sup>1</sup> state that standard anxiety management procedures are not particularly complicated; however, they caution that they

should only be used by SLPs [*speech-language pathologists*] who have had appropriate experience and/or training during their professional preparation and/or at some later stage, and that their use should be in accordance with the code of ethics of the individual SLP's [*speech-language pathologist's*] professional body. (p. 195–196)<sup>1</sup>

A report of a 2019 European conference about stuttering treatment,<sup>3</sup> with delegates from 29 countries, led the convenors to conclude that “there was unanimous agreement that SLPs are ideal personnel to provide basic CBT services for persistent stuttering” (p. 10). However, they cautioned that

... SLPs [*speech-language pathologists*] of today have a frontier to negotiate. They cannot function without being informed by the field of clinical psychology and, it seems, without ready access to a clinical psychologist when assessing and treating children, adolescents, and adults with persistent stuttering who experience anxiety. (p. 10)<sup>3</sup>

A contrasting view was expressed by a discussant at a 2022 clinical symposium about mental health and early stuttering.<sup>4</sup> The discussant stated that, for pre-school children who require anxiety intervention, referral to a clinical psychologist should not be necessary.

### ANXIETY MEASUREMENT FOR SPEECH-LANGUAGE PATHOLOGISTS

#### Background

It is important to stress that, for clinical psychologists and psychiatrists, detecting an anxiety problem and giving it a DSM-5 diagnosis is not simply a matter of administering formal assessments. Clinical psychologists and psychiatrists typically diagnose an anxiety disorder after a period during which they formally test, interview, and generally become familiar with a client. Such an assessment process would cover domains in addition to anxiety that are related to it, such as depression and stress. And psychological assessment of childhood anxiety is different to measuring the anxiety of adults. One difference is the possible limitation of child report about anxiety. Because of this, it is generally agreed that parent reports are essential input for assessing anxiety with children. Clinical psychologists administer tests, observe children, and interview them and their parents—and sometimes teachers—to form a diagnosis.

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<sup>†</sup> Thanks to Ross Menzies and Lisa Iverach for guidance with this material.

As noted during the previous lecture, adults seeking treatment for stuttering are often affected by social anxiety disorder. An overview of clinical measures for that specific disorder that clinical psychologists may use is available.<sup>5</sup> The following measures for social anxiety are suitable for administration by speech-language pathologists; they require no formal psychology qualifications to administer. However, they are not diagnostic tools for mental health disorders. An additional caution is warranted: some of these tools have been developed by clinical psychologists to assess anxiety without specific reference to stuttering. As such, it would be wise for speech-language pathologists to use these tests only for screening to determine the need for referral to a clinical psychologist. In what follows, each test is labelled as whether it was developed specifically about stuttering for use by speech-language pathologists, or whether it was developed by clinical psychologists about anxiety in the general population.

**The Unhelpful Thoughts and Beliefs About Stuttering (UTBAS) scales**

These scales were developed specifically about stuttering for use by speech-language pathologists.

*Overview*

The UTBAS scales provide a stuttering-specific measure of the unhelpful thoughts and beliefs that may drive social anxiety for those who stutter. The scale can be downloaded from the website of the Australian Stuttering Research Centre, along with translations into several languages.<sup>6</sup> An Italian<sup>7</sup> Japanese<sup>8</sup> and a Turkish<sup>9</sup> version of the scale have been developed. The UTBAS scales relate well to the OASES (see Lecture Four).<sup>10</sup>

Clinical psychologists and speech-language pathologists developed the scale<sup>11,12</sup> by producing a list of 66 commonly occurring unhelpful thoughts about stuttering expressed by those who stutter. The client indicates how frequently each thought occurs using a scale of 1 to 5, where 1 = *never or not at all*, 2 = *rarely or a little*, 3 = *sometimes or somewhat*, 4 = *often or a lot*, 5 = *always or totally*. The numerical scores are added to obtain a total score between 66 and 330.

To supplement this basic scale, there are two other scales that measure how much clients believe each thought, and how anxious each thought makes them feel. The three UTBAS scales are referred to as UTBAS I, II, and III. It is an option to give all three scales and combine the scores for a total UTBAS score between 198 and 990.

*Interpreting UTBAS scores*

The table below shows means and standard deviations for UTBAS I, II and III, and the total UTBAS score for 140 adult stuttering participants.<sup>12</sup> Of these participants, 24% met 12-month criteria for a DSM-IV social anxiety disorder diagnosis.

UTBAS I		UTBAS II		UTBAS III		TOTAL	
MEAN	SD	MEAN	SD	MEAN	SD	MEAN	SD
165	(52)	145	(53)	159	(62)	469	(160)

*A guide for clinical interview*

Arguably, the UTBAS is of most value as a guide for questions during an interview to establish whether a client’s speech anxiety might be clinically troublesome. For example, clinicians could adapt certain scale items to ask a client during an interview, “Have you ever thought that people would doubt your



ability because you stutter?" (Item 1), "Do you ever have the feeling that people are focusing on every word you say?" (Item 8), "Do you ever think that your stuttering will prevent you from being successful?" (Item 15), or "Have you ever thought that most people view those who stutter as less capable?" (Item 34).

Based on the responses obtained, and based also on general questioning to determine whether the client might be anxious about speech, clinicians may wish to use the UTBAS and other anxiety measures to provide a quantitative indication of the client's anxiety.

#### *Age range*

The UTBAS scales were developed for adults, but can be adapted for use with adolescents, with some minor wording changes to suit that age group. "I'm of no use in the workplace" is replaced with "I'm of no use in the classroom," and "I can't speak to people I find sexually attractive" is replaced with "I can't speak to people I find attractive." With adolescents, perhaps 16- and 17-year-olds, clinicians might be reasonably comfortable using the UTBAS means and standard deviations. This may not be so advisable with younger adolescents, and the scale is probably of limited use with school-age children.

#### *The UTBAS-6*

There is a six-item screening version of the UTBAS, known as the UTBAS-6,<sup>13</sup> which is presented in Appendix One to this lecture, as well as being downloadable from the website of the Australian Stuttering Research Centre.<sup>6</sup> This scale is recommended for routine clinical assessment of anxiety by generalist speech-language pathologists; the full UTBAS scales are more suitable for in-depth assessment by speech-language pathologists who specialise in stuttering and anxiety.

The six items can accurately reproduce the total score for each of the three subscales. The researchers who developed the scale recommend that when the total UTBAS-6 score falls in or above the fifth decile, the client should be referred for psychological assessment. However, this does not mean that a score below the fifth decile excludes a clinically significant anxiety problem. The researchers indicate that "the decision about referral to a psychologist will be based on a combination of UTBAS-6 scores, any other clinical measures, and clinical judgement" (p. 970).<sup>13</sup>

These are the test items of the UTBAS-6, covering negative thoughts in the domains of fear of negative evaluation (1–2), avoidance (3), self-doubt and lack of confidence (4), and hopelessness (5–6):

- (1) People will think I'm strange.
- (2) People will think I'm incompetent because I stutter.
- (3) I don't want to go—people won't like me.
- (4) I'll never finish explaining my point—they'll misunderstand me.
- (5) What's the point of even trying to speak—it never comes out right.
- (6) I'll never be successful because of my stutter.

Appendix Two to this lecture shows an example of an UTBAS-6 form completed by a client with clinically significant anxiety.

#### *The Safety Behaviour Checklist*

This scale was developed specifically about stuttering for use by speech-language pathologists.

As noted in Lecture Ten, safety behaviours are used by those who are socially anxious as an attempt to prevent a perceived threat or negative event from occurring. They prevent fear extinction and may even cause a feared social outcome to occur. The Safety Behaviour Checklist is publicly available,<sup>14</sup> and, as is the case with the UTBAS, it is useful to guide a clinical interview. As outlined in Lecture Ten (see *Safety behaviours during stuttering treatment*), there are five categories of safety behaviours in this checklist: General Safety Behaviours, Practice and Rehearsal, General Avoidance, Choose Safe and Easy People, and Control Related. The Safety Behaviours Checklist has been translated into Persian,<sup>15</sup> and its validity and reliability in that language were established.

## The Fear of Negative Evaluation (FNE) scale

The various versions of this test were developed by clinical psychologists about anxiety in the general population; they were not developed specifically about stuttering.

### *The original 30-item FNE scale*

The FNE scale was originally published in 1969<sup>16</sup> as a 30-item self-report questionnaire where respondents indicate true or false to statements referring to the expectation and fear of negative evaluation from others. For responses that suggest social anxiety, one point is scored, and for responses that suggest no social anxiety, no point is scored. There are several reports published with FNE data for stuttering participants.<sup>12,17</sup>

### *The Brief FNE scale*

After the popularity of the 30-item version of the scale, several publications focused on abbreviating the measure to either a 12-item version or an 8-item version.<sup>18</sup> A general conclusion from this research is that the 8-item version is useable because it has similar properties to the original 30-item scale. The 8-item version is generally referred to as the BFNE-S. The scale has been translated, and its reliability and validity established, with Vietnamese adults who stutter.<sup>19</sup>

Each of the eight items is scored on a scale of 0–4, where 0 = *not at all characteristic of me*, 1 = *a little characteristic of me*, 2 = *somewhat characteristic of me*, 3 = *very characteristic of me*, 4 = *entirely characteristic of me*. Numerical scores of each item are then summed to give a total score. The range of scores will therefore be 0–32.

These are the test items of the BFNE-S:

- (1) I worry about what other people will think of me even when I know it doesn't make any difference.
- (2) I am frequently afraid of other people noticing my shortcomings.
- (3) I am afraid that others will not approve of me.
- (4) I am afraid that other people will find fault with me.
- (5) When I am talking to someone, I worry about what they may be thinking about me.
- (6) I am usually worried about what kind of impression I make.
- (7) Sometimes I think I am too concerned with what other people think of me.
- (8) I often worry that I will say or do wrong things.

### *Age range*

The 30-item FNE and the Brief FNE scales were developed for adults and have not been adapted for younger clients. So, the advice for using them with adolescents is essentially similar to that for the UTBAS. For older adolescents, age 16 or 17 years, it may be reasonable to use the norms that are available for adults, and the test items can be useful to guide a clinical interview about anxiety. However, it would be incautious to apply the available norms to younger adolescents or school-age children.

### *Interpreting Brief FNE scores*

*Sensitivity* is the true positive rate and *specificity* is the true negative rate (see Lecture Two). One report<sup>20</sup> shows Brief FNE sensitivity and specificity values for identifying people with social anxiety disorder (Table 3, p. 826). Based on achieving an ideal trade-off between sensitivity and specificity, the table suggests a cut-off score of 25 for potentially clinically significant anxiety. This score provides sensitivity around 65% and specificity around 80%. In other words, a score of 25 gives a 65% chance of indicating a problem when there is one and a 20% chance of indicating a problem when there is not one.

The report<sup>20</sup> is probably worth reading prior to using the Brief FNE during clinical practice to screen for clinically significant anxiety. The authors point out that the user can consult Table 3 (p. 826)<sup>20</sup> to

form a cut-off score according to individual need. So, for example, if a clinician wanted to identify as many clients as possible with clinically significant anxiety, and were not particularly concerned about making a mistake, a cut-off score of 15 might be used. That would give sensitivity of around 90% but specificity—an error rate—of around 40%. Screening always involves such a trade-off between sensitivity and specificity.

### Subjective Units of Distress Scale (SUDS)

This test was developed by clinical psychologists about anxiety in the general population; it was not developed specifically about stuttering. However, its use by speech-language pathologists in the context of stuttering treatment is normally straightforward.

Previous measures discussed have dealt with *trait anxiety*, which is anxiety linked to temperament. However, the SUDS measures state anxiety, which is an immediate emotional response to everyday experiences. The SUDS is usually attributed to the psychologist Wolpe during the 1960s.<sup>21</sup> Clinical psychologists today commonly use this scale to evaluate the distress experienced at a particular time or during a particular situation, or to predict the level of distress for any coming situation. As such, it can be useful for state anxiety assessment during stuttering treatment for adults and adolescents.

Ratings can be made on an 11-point scale from 0–10 or a 101-point scale from 0–100. In either case, 0 = *no anxiety* and 10 or 100 = *extreme anxiety*. The SUDS is quick to administer and can be used by clients for self-assessment during everyday speaking situations. The scale has been shown to be valid.<sup>22</sup>

For clinical purposes it would be appropriate for a speech-language pathologist to use the SUDS during treatment to determine the level of client anxiety experienced during speaking situations, or when thinking about those situations. This could provide information about whether stuttering reductions in those situations are associated with anxiety reductions, or whether anxiety treatment is needed in addition to speech treatment. The 11-point version of the scale is presented in the diagram.



### Communication Attitude Tests: CAT, KiddyCat, BigCat,

These tests were developed specifically about stuttering for use by speech-language pathologists.

There is a long history in the field about measurement of a dimension referred to as communication attitude. The idea of measuring such a construct can be linked, according to one account,<sup>23</sup> to the Anticipatory Struggle Hypothesis, which is an outdated causal perspective about stuttering, mentioned during Lecture Three. The Anticipatory Struggle Hypothesis is summarised in a prominent textbook<sup>24</sup> (pp. 149–151). It posits that stuttering is driven and sustained by an early developing belief that speech is difficult. Two seminal developments in measuring such a construct in adults occurred during the mid-20<sup>th</sup> Century.<sup>25,26</sup> At present, there are three commercially available tests to measure the construct of communication attitude, spanning the ages of pre-school, school age and adults. On the face of it, for those who stutter, it might be anticipated that dimensions such as a negative attitude to communication and social anxiety have some overlap. As indicated in the following, some test items confirm that impression.





### *The Communication Attitude Test (CAT)*

The Communication Attitude Test,<sup>27</sup> often referred to as CAT, is designed specifically for children who stutter who are 6–15 years old. It has been available for some time.<sup>23</sup> It requires children to respond *true* or *false* to 35 questions. For children who stutter, it might be anticipated that there is some overlap of the construct “communication attitude” with anxiety. Some test items confirm that suggestion: for example, “my classmates ... think I talk funny,” “some kids make fun of the way I talk,” (p. 73), and “I ... worry about the way I talk.”<sup>28</sup> The Communication Attitude Test has been shown, in many cultures and languages, to distinguish between children who stutter and controls, as reviewed during Lecture Eleven.

A systematic review dealing with measures of the psychological impact of stuttering on school-age children<sup>29</sup> included the Communication Attitude Test. The authors reported that there were few well-developed measures for this age group, and noted the problematic nature of this situation; psychological issues have been documented to emerge for stuttering children during this time of life (see Lecture Eleven). According to that systematic review, compared to other tests of psychological impact of stuttering, the Communication Attitude test had the most comprehensive support for its measurement properties.<sup>†</sup> The authors noted that there was no available evidence for a change of the test scores after clinical intervention. That observation is consistent with another systematic review<sup>30</sup> that identified six clinical reports that presented, for 6–12-year-olds, pre-treatment and post-treatment data for the Communication Attitude Test. No evidence was reported for any post-treatment improvement.

The authors of the first review<sup>29</sup> found that few measures of psychological impact of stuttering for school-age children had their developmental data published in peer-reviewed journals. Consequently, they included the Communication Attitude test in the report, even though the only supportive data for it are reported in the commercially available test manual. This is a caveat for the use of the test: its supportive data are not peer reviewed and are only available for scrutiny when the user has purchased the manual.

### *The KiddyCat*

This is a version of the Communication Attitude Test for children younger than 6 years.<sup>31</sup> As is the case for the Communication Attitude Test, as outlined earlier, its items suggest that it has some overlap with social anxiety. Children are asked 12 *yes/no* questions, such as “is it hard for you to say your name,” “do your words come out easily,” “do mom and dad like how you talk,” and “do people like how you talk” (p. 229).<sup>31</sup> As noted in Lecture 10, there are several reports of the KiddyCat with pre-schoolers who stutter and control children.

### *The BigCat*

This is an adult version of the Communication Attitude Test,<sup>32</sup> comprising 34 items, to each of which respondents indicate whether it is “mostly true” or “mostly false.” The reliability and validity of the test, including its capacity to distinguish between adults who stutter and controls, has been demonstrated in participants from the United States,<sup>33,34</sup> Poland,<sup>35</sup> Iran,<sup>36</sup> and Kannada-speaking India.<sup>37</sup> As is the case for the Communication Attitude Test and the KiddyCat, the BigCat appears to have some relation to speech-related anxiety: “My speech is as good as that of most people,” “I will usually have some trouble with my speech,” “my speech does not affect the way I interact with people,” and “I am self conscious about the way I speak” (p. 202).<sup>33</sup>

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<sup>†</sup> The authors also cited the OASES for school-age children (see Lecture Four) as having empirical support of its development.

## The Spence Children's Anxiety Scale

The various versions of this test were developed by clinical psychologists about anxiety in the general population; they were not developed specifically about stuttering.

### Overview

The Spence Children's Anxiety Scale has been shown to be reliable and valid, and it has comprehensive normative data available. The scale is well established, extensively used, accessible from a website without charge,<sup>38</sup> and is available in many languages.

### Child and parent version (8–15 years)

There are child and parent response versions for this age group,<sup>38</sup> the child version containing 45 items and the parent version containing 38 items. Examples of items are "my child worries about things," "my child is afraid of the dark," "my child complains of feeling afraid," and "my child worries about being away from us/me." Responses to items are scored using a four point scale, where 0 = *never*, 1 = *sometimes*, 2 = *often*, 3 = *always*. Individual item scores are used to calculate sub-scale scores for various domains of anxiety: generalized anxiety, panic/agoraphobia, social phobia, separation anxiety, obsessive compulsive disorder, and physical injury fears.

T-scores are available for the raw test scores. T-scores are rescaled so that the distribution has a mean of 50 and a standard deviation of 10. This enables comparison of results across the six subscales. A score of less than 10 above the mean is not considered to be concerning. The scale is not intended as a standalone diagnostic instrument:

The SCAS is not intended as a diagnostic instrument when used in isolation. Rather it is designed to provide an indication of the nature and extent of anxiety symptoms to assist in the diagnostic process. It is recommended that clinicians use the scale in partnership with a structured clinical interview.<sup>38</sup>

However, speech-language pathologists could use the parent report scale for screening purposes to determine any need for a clinical psychology referral. In which case, it is not advisable for a speech-language pathologist to use the subscales for screening purposes, only the total score.

### Pre-school version (3–5 years)

The scales have a version for 3–5-year-olds<sup>38</sup> comprising 28 parent report items that provide an overall anxiety measure. Parents follow instructions, which can be downloaded from the website.<sup>38</sup> Examples of test items are "has difficulty stopping himself/herself from worrying," "is scared of heights (high places)," and "is afraid of crowded or closed-in spaces." Parents respond to each item using a 5-point scale to indicate the extent to which each of the 28 statements pertain to their children, where 0 = *not true at all*, 1 = *seldom true*, 2 = *sometimes true*, 3 = *quite often true*, and 4 = *very often true*. Individual item scores are used to calculate sub-scale scores for specific aspects of child anxiety: generalized anxiety, social anxiety, obsessive compulsive disorder, physical injury fears, and separation anxiety. A total numerical score is obtained from those responses. T-scores are available for the subscales and the total score. As is the case with the school-age version, it is advisable for speech-language pathologists to use only the total scores for screening.

## The Preschool Anxiety Scale Revised

This test was developed by clinical psychologists about anxiety in the general population; it was not developed specifically about stuttering.

The Preschool Anxiety Scale Revised<sup>39</sup> is a parent-report anxiety measure of anxiety for children younger than 6 years. It consists of 28 items. For each item, parents select the response that best describes the child, using a 5-point scale, where 0 = *not at all true*, 1 = *seldom true*, 2 = *sometimes true*, 3 = *quite often true*, and 4 = *very often true*. A total score is obtained by adding scores for all items, with a maximum total score of 112. Individual item scores are used to calculate sub-scale scores for four categories: social anxiety ("worries that s/he will do something to look stupid in front of other people"), generalized anxiety ("has difficulty stopping him/herself from worrying"),

separation anxiety (“would be upset at sleeping away from home”), and specific fears (“is afraid of insect and/or spiders”).

The Preschool Anxiety Scale Revised is publicly available in English and seven other languages. Normative data are provided<sup>39</sup> for 764 mothers and 418 fathers of Australian 3–5 year old children. Cut-off scores are not provided for determining whether a child is in the clinical range. However, Table 3 (p. 405)<sup>39</sup> provides mean total and subscale scores based on mother and father report. Elevated scores compared to those means can be used to assist clinical judgement about the need for referral to a clinical psychologist.

## EVIDENCE-BASED ANXIETY TREATMENT FOR STUTTERING

### Cognitive Behaviour Therapy (CBT)

Cognitive Behaviour Therapy, known generally as CBT, is the flagship clinical psychology intervention for a range of psychological problems involving negative emotions such as anxiety, depression, and anger. It has been shown efficacious with a range of DSM-5 disorders. A search of the Web of Science database<sup>40</sup> shows thousands of publications dealing with the method. A tutorial<sup>1</sup> about CBT for anxiety with stuttering gives an overview of the four standard components of the treatment, with specific reference to stuttering: exposure, behavioural experiments, cognitive restructuring, and attentional training. An overview of Cognitive Behaviour Therapy is given on the Australian Association for Cognitive and Behaviour Therapy website.<sup>41</sup>

There is evidence from the 1970s that anxiety treatments such as desensitisation and meditation may benefit those who stutter,<sup>42,43</sup> and it is common to incorporate CBT, or components of it, within speech restructuring treatments.<sup>44</sup> There has been a report about the effects of exposure therapy on stuttering<sup>45</sup> using six participants in a multiple baseline design experiment. The exposure task was 10 sessions of speaking to an audience. Before the experiment, all six participants met criteria for diagnosis of social anxiety disorder, and after the experiment only one retained that diagnosis. However, the results are difficult to interpret because the participants also received varying sessions of progressive muscular relaxation, ranging from none to four sessions.

There have been only two conference reports about the value of an entire CBT treatment for those who stutter,<sup>46,47</sup> and only three published clinical trials that conform to the discipline standards for a clinical trial of stuttering treatment that were outlined during Lecture Five.

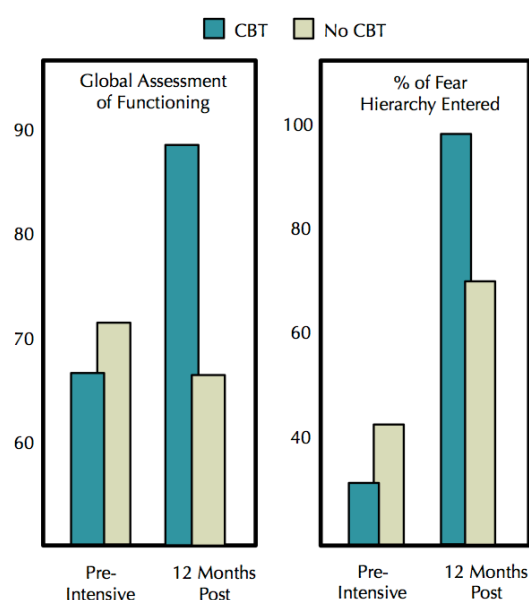
### CBT for stuttering: A clinical randomised controlled trial

#### Design

The first trial<sup>48</sup> involved 32 stuttering participants, 60% of whom were diagnosed with social anxiety disorder. They were recruited to a randomised controlled trial of CBT adapted for the needs of stuttering clients. Participants were randomly allocated to receive a CBT package followed by an intensive speech restructuring treatment, or an intensive speech restructuring treatment alone. A clinical psychologist gave the CBT treatments, which were a standard 15 hours, in weekly sessions for 10 weeks. Seven sessions were 1 hour, one was 2 hours, and two were 3 hours. There were seven participant dropouts (22%).

#### Results

Results are presented in the figure. The trial clearly showed that the addition of CBT to speech



restructuring treatment did not reduce %SS scores at all. However, the trial showed no social anxiety disorder diagnoses, and an overall general improvement of psychological functioning, after CBT. Immediately after CBT, statistically and clinically significant improvements were reported for Global Assessment of Functioning. The DSM-IV index is a score out of 100 indicating general mental health and wellbeing, quality of functioning during daily life free of psychiatric difficulties, and engagement with the world. A clinical psychologist or a psychiatrist gives the score after a full diagnostic interview. Similar statistically and clinically significant results were found immediately after CBT for the 30-item FNE Scale, UTBAS scores, and the Social Phobia and Anxiety Inventory.<sup>49</sup>

At 12 months post-treatment, a statistically significant result for Global Assessment of Functioning scores remained. There was little change for participants in the control arm, but the participants in the CBT arm were in the typical range of psychological functioning post-treatment. Additionally, participants assembled a hierarchy of their least feared to their most feared speaking situation. At 12 months post-treatment, participants in the CBT arm were able, on average, to enter almost 100% of their fear hierarchies. The control arm showed improvement of this measure after speech treatment, but not to the same extent as the participants in the CBT arm. The difference between the groups was significant at 12 months post-treatment.

### **iGlebe: A standalone Internet CBT for stuttering**

#### *Clinical issues driving the development*

Despite its promise, the clinical trial just described raises various clinical issues that researchers have noted.<sup>50</sup> Most obviously, a clinical psychologist gave the treatment. As discussed earlier, it may be appropriate for speech-language pathologists to provide CBT to clients who stutter if they have the appropriate professional preparation and training. However, this raises the matter of whether such speech-language pathology interventions would be as effective as those provided by clinical psychologists. It would be difficult to argue that this would be the case.

Another issue is the limited viability for every speech-language pathologist who manages stuttering caseloads to attain appropriate professional preparation for managing anxiety. Ideally, that preparation would involve formal postgraduate CBT qualifications, which is not a foreseeable prospect for all speech-language pathologists who routinely manage stuttering caseloads. Nor is it a foreseeable prospect that speech-language pathology professional preparation programs worldwide will universally provide training to benchmark clinical psychology standards for CBT treatment. Nor is it viable that, in every case, clients who require stuttering and anxiety management would receive these services concurrently from a speech-language pathologist and a clinical psychologist.

#### *Standalone Internet CBT as a solution*

A potential solution to those problems is a standalone Internet-based CBT treatment for stuttering clients,<sup>50</sup> in other words, an Internet treatment program that does not require clients to have personal contact with a clinician. This is a common approach to mental health problems in clinical psychology.<sup>51</sup> It would enable speech-language pathologists to integrate CBT treatment with speech restructuring treatment without needing psychological training or access to a clinical psychologist. They would be able to do so in a cost neutral manner.

A review<sup>52</sup> argued that Internet-based CBT treatment could fully replace a human therapist if treatments were customised to the individual user. Instead of a standard approach to treatment, the authors argued that an Internet-based treatment could begin with comprehensive assessment of individual client anxiety features, such as unhelpful thoughts and beliefs, as occurs with a standard clinic assessment. The authors also argued that human therapist simulation would require corrective feedback for incorrect client responses during the learning process of CBT, tracking of client access to the program with encouragement for compliance, and reminders for failures to log on.

Additionally, treatment “dose” was raised as an issue with standalone Internet treatments; users require “a large number of opportunities to engage in cognitive and behavioural tasks relevant to their problems” (p. 251).<sup>52</sup> Finally, the authors argued that the design of a successful internet-based CBT

treatment would need to incorporate features that simulate contact with a human therapist as much as possible.

### Program design

*Background.* Incorporating the foregoing guidelines, the authors designed a standalone Internet CBT treatment: iGlebe.<sup>† 53</sup> The treatment is available at no cost to the public.<sup>54</sup> The program design is based on the original clinic treatment<sup>48</sup> and incorporates components of the Clark and Wells social anxiety model described during Lecture Ten.<sup>55,56</sup> The program incorporates the faces and voices of a male and female clinical psychologist who communicate to the user throughout the treatment. The treatment is designed around an on-line pre-treatment assessment, conducted within the iGlebe program, which includes the 30-item FNE, UTBAS, and the Depression Anxiety Stress Scales.<sup>57</sup> A Stuttering Specific Avoidance Scale was developed specifically for the program. It presents the user with 55 common daily life situations and requires each of the situations to be scored on a five-point scale where 1 = *never avoid* and 5 = *always avoid*. When users have completed the treatment, they repeat these online assessments.

*Section One.* Section One introduces the user to the voices and images of the two clinical psychologists who present iGlebe. The program explains to the user the cognitive model of emotion, involving the relationship between events, thoughts, and emotions: events prompt thoughts, which prompt emotions. Examples are provided of how people can control their thoughts, and hence control emotions. Users are shown how both negative and positive thoughts and emotions may emerge from the same event. An example provided (see the figure) is of missing a bus because it departs ahead of schedule. The figure shows how this event may promote thoughts that lead either to positive or negative emotions.

The iGlebe clinical psychologist then presents a “thinking exercise” where the user is presented with a life situation that does not involve speech. Following this, users are required to write different thoughts about the situation that could lead to emotions of anxiety, anger, sadness, and happiness. The program then compares the user responses with the responses prepared by the clinical psychologist.

Next, the program presents a scenario where a woman asks a sales assistant for something, stuttering while doing so, and the sales assistant asks her to repeat the request. The user is required to repeat the thinking exercise and responses are again compared to the iGlebe responses. Of the given responses that would lead to anxiety is “the sales assistant thinks I am stupid,” one that would lead to anger is “she has no right to treat me this way,” one that would lead to sadness is “I’m hopeless, I can’t do anything right,” and a response that would lead to happiness is “I

iGlebe  
A social anxiety treatment  
for adults who stutter



<sup>†</sup> In early publications the program was referred to as CBTPsych.

only had to repeat once what I was asking for.” Similar thinking exercises recur throughout Section One.

iGlebe then shifts from how thoughts in response to events cause emotions, to the idea of causal thoughts: an idea or belief that of itself would cause an emotion. The program then links into the three assumptions that underlie the Clarke and Wells model of social anxiety: excessively high standards of social performance, beliefs about performing in a certain way in social situations, and unconditional negative self-beliefs. iGlebe presents examples of such causal thoughts.

The program continues with an explanation of common *cognitive errors*, otherwise known as *cognitive distortions*, such as those outlined in the following table. These are typically incorporated within CBT.<sup>58</sup> iGlebe presents numerous examples of these cognitive errors, such as “I am going to make a fool of myself at the party” and “I have to look fantastic all of the time” and the user is required to identify which of the cognitive errors they are.

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MIND READING	<i>Assuming people are thinking negative thoughts about you when there is no real evidence that they are.</i>
FORTUNE TELLING	<i>Arbitrarily predicting that things will turn out badly.</i>
EMOTIONAL REASONING	<i>The way you feel about yourself is reality: “I feel stupid so I must be stupid.”</i>
MENTAL FILTERING	<i>Dwelling on the negatives and discounting the positives.</i>
“SHOULD” THINKING	<i>Developing negative emotions about yourself and others based on internalised rules about the behaviours of others: “That shopkeeper should not have been rude.”</i>
OVERGENERALISATION	<i>Interpreting negative events as part of a never ending pattern of defeat.</i>
ALL OR NOTHING THINKING	<i>Thinking in black and white categories with nothing between: “My spouse and I disagree on some things so we have a poor relationship.”</i>
DISCOUNTING THE POSITIVES	<i>Belief that positive achievements don’t count in evaluating yourself.</i>
LABELLING	<i>Thinking “I am a loser” instead of “I made a mistake.”</i>
PERSONALISATION	<i>Blaming yourself for something that was not entirely your fault.</i>
BLAME	<i>Overlooking how your attitude and behaviour may contribute to problems.</i>

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*Section Two.* This section uses the online UTBAS user scores from the pre-treatment assessment to create an individualised profile of unhelpful thoughts and beliefs about stuttering. It is a standard CBT technique to challenge such unhelpful thoughts that may cause negative emotions, which in this case is anxiety. These standard cognitive challenges are: (pp. 264–265)<sup>53</sup>

- (1) What evidence do you have for the thought?
- (2) What evidence do you have against the thought?
- (3) What would you tell a friend, to help, if he/she had the thought?
- (4) Think of your calmest, most rational and supportive friend or family member. How would he/she react to the causal thought? What would he/she say?
- (5) Are you worrying about an outcome you can't control? Is there any point to this type of worry?

- (6) What does the thought do for you? How does it make you feel? Is it helpful or just distressing?
- (7) What good things would you gain if you gave up the thought? How would your life be different if you didn't believe the thought?
- (8) If the causal thought was true, what is the worst outcome? Is it as bad as you think?

There are 66 UTBAS items. For all these items, iGlebe has cognitive restructuring sample answers for each of the eight probe questions in the above table, totalling 528 sample answers. To ensure an adequate dose of cognitive restructuring, iGlebe requires users to write at least 40 different restructurings of their unhelpful thoughts and beliefs. For each cognitive challenge that the user writes, iGlebe provides a predetermined challenge from its database for comparison. For example, in response to the unhelpful thought "It's impossible to be successful if you stutter," the following was a user's response to the probe question "what would you tell a friend, to help, if he/she had the thought?" (p. 264):

It is absolutely wrong. You can be successful in many things despite you [sic] stutter, look at your past performance, you succeed sometime.<sup>53</sup>

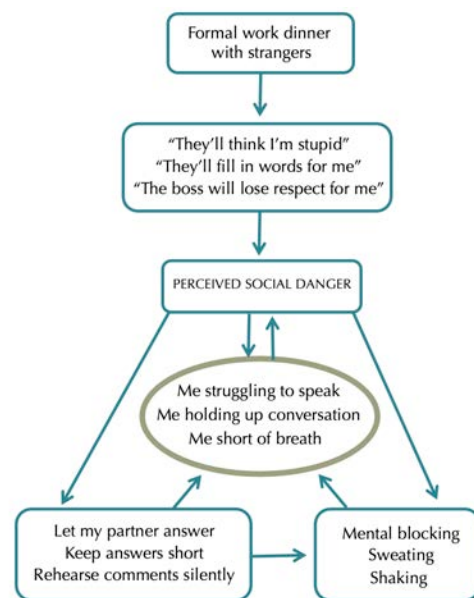
The iGlebe sample answer was:

Don't be silly! Lots of people who stutter are successful. This thought is so self-defeating. You need to beat it!<sup>53</sup>

*Section Three.* This section is an extensive psychoeducation package based on the Clark and Wells model. Situation avoidance and safety behaviours are explained in detail, with examples. Users are then guided to build an individualised model of their anxiety, incorporating information from their pre-treatment assessments of unhelpful thoughts and beliefs and avoided situations. The guide includes avoided situations, thoughts that drive anxiety and avoidance, safety behaviours, mental self-images, and physical anxiety symptoms. When the user has constructed the model, it is used later in the program to establish behavioural experiments and to target unhelpful imagery for correction. The diagram gives an example of an individual formulation that might occur for a user.\* It relates to the social event of a formal work dinner with strangers.

The impending social event activates the assumptions outlined earlier: excessively high standards of social performance, beliefs about performing in a certain way in social situations, and unconditional negative self-beliefs. These assumptions activate causal thoughts for anxiety such as "they'll think I'm stupid," "they will humiliate me by filling in words when I have a speech block," and "the boss will lose respect for me when I make a fool of myself in front of his colleagues." Such thoughts lead to a perception that the impending social event is dangerous. Safety behaviours are planned for during the dinner to avoid the feared outcomes: letting a partner do most of the talking, keeping any answers to questions short, and silently rehearsing every utterance before saying it.

During the dinner, negative self-focus includes images of struggling to speak, holding up the conversation with stuttering, and being short of breath. These are compounded by a destructive cycle where safety behaviours have the reverse effect to what is intended by making the speaker appear odd, unfriendly, distant, or aloof, which worsens feelings of self-



\* Adapted and reproduced with permission: Clark, D. M. (2001). A cognitive perspective on social phobia. In W. R. Crozier & L. E. Alden (Eds.), *International handbook of social anxiety: Concepts, research and interventions relating to the self and shyness*, (p. 405–430), Hoboken, NJ: Wiley. © 2001 John Wiley & Sons, Ltd.

consciousness, thereby feeding into a cycle involving negative self-processing and anxiety symptoms of mental blocking, sweating, and shaking during the dinner.

Then, after the dinner there is rumination about how humiliating the whole event was and how awkward it would be speaking to the boss at work the next day. All that can be recalled about the dinner is struggling to speak, holding up everyone's conversation, and gasping for breath while trying to speak. The dinner is added to a mental list of previous failures and confirms the expectation that such events in the future will be similar.

*Section Four.* This section presents behavioural experiments about feared situations. One of the iGlebe clinical psychologists says this to the users:

"This is a particularly important component of the treatment package, because it introduces you to behavioural experiments. In previous sections of the program, you've learned that social anxiety is driven by negative thoughts and maintained by safety behaviours and avoidance. Behavioural experiments are a way to test out your thoughts or your predications about situations and they're fairly straightforward really to understand. You make a prediction about what will happen in a particular social situation. You enter the social situation and engage in a real way and you discover whether your prediction comes true or not."

The success of the technique relies on the fact that those who are socially anxious typically overestimate the likelihood and seriousness of a predicted negative outcome.

For behavioural experiments, iGlebe uses the avoided situations that each user recorded with the Stuttering Specific Avoidance Scale at the pre-treatment online assessment. Around 10 behavioural experiments are designed for each user. A list of 21 common predictions for those who stutter is presented, from which the user selects three. Examples of such predictions are "people will walk away," "they will not talk to me," and "I will forget what I am going to say." For each prediction the user uses a 100-point scale to indicate the perceived probability of its occurrence. iGlebe leads users to compare the actual outcome with the predicted outcome. Users are instructed to repeat behavioural experiments until they are no longer anxious in the avoided situations.

iGlebe has the capacity to create 3,620 different behavioural experiments for users based on their pre-treatment assessment data. Users are instructed to carefully avoid using their typical safety behaviours, as identified in Section Three, during behavioural experiments. The following is an example of a behavioural experiment provided by iGlebe and how it turned out.

A man has avoided going into banks because of a fear that he would not be able to make his needs known to the teller and that the teller would be condescending to him. His task was to go into a bank and make a deposit into his account. When he entered the bank, he was anxious, and more so when he approached the teller. The teller greeted him in a friendly manner and asked how she could help. He stuttered a few times, but nonetheless was able to communicate his request. The teller made the deposit and courteously wished him a good day, without any sign of condescension. So, the outcome was different from the prediction. Even if the teller did notice the man's stuttering, it did not interfere with him achieving the purpose of going to the bank and it did not prompt any condescending behaviour.

*Section Five.* This section continues material from Section Two, which challenges fear of negative evaluation. Users are guided, by means of a sample essay, to write about "why it doesn't matter what other people think of me." One of the iGlebe clinical psychologists says the following as part of the preparation for this exercise:

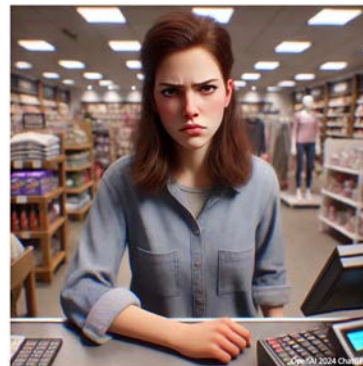
"When you care about the opinions that others hold of you, you're giving them tremendous power over your emotional life. You're saying in a sense that you can only be happy if they're happy with you, you can only feel good if they feel good about you. Giving somebody that much power over your sense of self worth doesn't make any sense if you really think about it."

The second part of this section targets unhelpful "should" cognitions, and the problems they cause. "Should" cognitions refers to internalised rules people have about the behaviours of others and the



tendency to become angry if those rules are broken. The iGlebe psychologist informs users of the problems with becoming angry over something you think should not have happened but is now in the past and cannot be changed. Also, the user is informed that people cannot be prevented from behaving in ways you think they should not. Additionally, there are many different perspectives about how people “should” behave, and it is irrational to think that yours is the correct one.

iGlebe provides the example of a man stuttering severely with a shop assistant who said, “hurry up, there is a long cue.” This made him angry and affected his mood for the rest of the day and could well have made him anxious about future dealings with shop assistants. He kept thinking “she shouldn’t have been so rude.” But there is no point in that thinking because it can’t change what happened. A better outcome for him would have been just to accept that the shop assistant was tired, rude, and insensitive, and too young to know any better, and for him to move on and not waste mental energy and emotions on someone he found to be so distasteful. The iGlebe psychologist points out that there are many different perspectives about this scenario and the man’s perspective is just one of them.



Other perspectives are that the shop assistant was being considerate to other shoppers, that she had the right to do her job as she sees fit, and that she had the right to free speech. There is nothing innately correct about the man’s view that “she shouldn’t have been so rude.”

iGlebe targets maladaptive “should” thinking by requiring users to select three of 17 “should” cognitions commonly associated with anxiety. For each of the three selected “should” statements, the program guides the user to explore the advantages and disadvantages of each. Users are then required to choose three of the “should” cognitions and to construct their own narrative for each of them. To assist the user, iGlebe provides 34 different sample responses for each of the common “should” cognitions.

*Section Six.* This section is designed to repair the imagery that leads to a negative self-focus during social situations and to establish a different perception of social encounters. There are four projected benefits from obtaining a healthy control of attention. First, it is intended that gaining control of imagery about social encounters will prevent the post-event rumination that can distress those with social anxiety and perpetuate the anxiety. Second, the problem of distorted observer perspective is targeted so that users obtain a correct picture of how people really respond to them in social situations. Third, attention will focus away from any negative events in a social encounter, which are likely to be minor, towards neutral or positive aspects of the encounter. Finally, it is projected that users will break their cycle of failing to disconfirm negative expectations during social encounters and will be able to find evidence that disconfirms those beliefs.

The first step is skills-based attentional training,<sup>59</sup> which first trains the user to control where attention rests at any moment, using the attentional training technique. The user downloads an audio file from iGlebe, in which the psychologist’s voice provides training in shifting attention rapidly from one focus to another. When the user has practised the attentional training technique daily for some weeks and mastered it, the iGlebe clinical psychologist introduces the situational attention refocusing technique:

“Situational attentional refocusing builds on your ability to place your attention where you wish. There is considerable research ... to suggest that anxious individuals place too much of their attention on negative aspects of social settings. I’m sure you’ve experienced this. Where attention seems caught by one negative person or one negative aspect of the environment, one person who you think is being critical of you. You don’t seem to be able to focus on anything else. Well we want you with your new attentional skills to enter social spaces in an unbiased way, moving your attention through the positive aspects of the situation.”

iGlebe then presents users with a list of their commonly avoided everyday situations, scored in the online pre-treatment assessment with the Stuttering Specific Avoidance Scale. The user is then

required to choose three of these situations and practise the situational attentional refocusing technique for each of them and to record what occurred during each practice. The user is urged to continually practise this technique.

The final part of Section Six deals with problems of mental imagery that involve the biased observer perspective, which often affects those with social anxiety, as discussed during Lecture Ten. The iGlebe clinical psychologist says to the user:

“Research has consistently shown that people with social anxiety lay down distorted images into their memory. Now this is very important because anxious individuals are basing a lot of their fear on going into social situations on their past memories on how they performed ... the memories that socially anxious individuals have include images of what they actually looked like in the events in which they were anxious ... they remember seeing themselves performing in the social event as if they had been an observer to the event. Now ... obviously these negative memories must be false; no one sees themselves when they speak.”

In this part, iGlebe presents an example of a social encounter during which stuttering occurs. The images of this encounter recur for the person two years later. The man recalls seeing himself looking anxious and tense during the event and those present evaluating him quizzically and apprehensively, and generally having a negative view of him, as shown in the image below on the left.



However, this recall of the situation cannot be correct and what actually occurred—the field perspective—was more like the image on the right. Yet, years later, he still ruminates about how badly he thought the social encounter went and the thought of it all makes him ruminate about how badly future social encounters will turn out how and makes him feel anxious about them before they even occur. The iGlebe program invites users to test whether this is an issue for them. The iGlebe clinical psychologist says to the user:

“Do you think you show this "observer" or "external camera" bias? You can test this out by simply closing your eyes and remembering images from past anxious speaking situations. Do you see yourself in the image, or are you simply seeing the faces of those around you? Remember, if your memory is displaying images of your own face, it is playing tricks on you! Such images simply cannot be accurate! Unless you are telling us that you were standing in front of a large mirror in these social events, you simply could not see yourself doing anything!”

The final part of Section Six deals with the “re-scripting” technique<sup>60</sup> for faulty images of past events such as in the previous example. Users download an audio file from iGlebe, in which a psychologist’s voice provides guidance to mentally go over a past event and re-script it so that it is different from the troublesome version. The mental image becomes one where the social event is going well, people are smiling and enjoying your company, and you are not stuttering. Users are instructed to repeat this exercise several times for each false and biased memory of a social event.

*Section Seven.* This final section deals with relapse prevention. It emphasises that minor setbacks are inevitable and should not be interpreted as relapse. The clinical psychologist guides users to recognise when they are vulnerable to anxiety setbacks, such as at times of stress or fatigue. The critical point is made that falling back into avoidance patterns never helps with anxiety setbacks; avoidance only perpetuates and worsens anxiety.

## A Phase II clinical trial of iGlebe

### Background

After two positive Phase I trials,<sup>53,50</sup> the developers conducted a Phase II trial of iGlebe.<sup>61</sup> The Phase I trials involved pre-treatment and post-treatment assessments at speech clinics. In effect, then, the trials were not standalone in the strictest sense, because such clinic contact may have been somehow associated with participant compliance. Hence the Phase II trial involved no direct participant contact of any kind from researchers or clinicians.

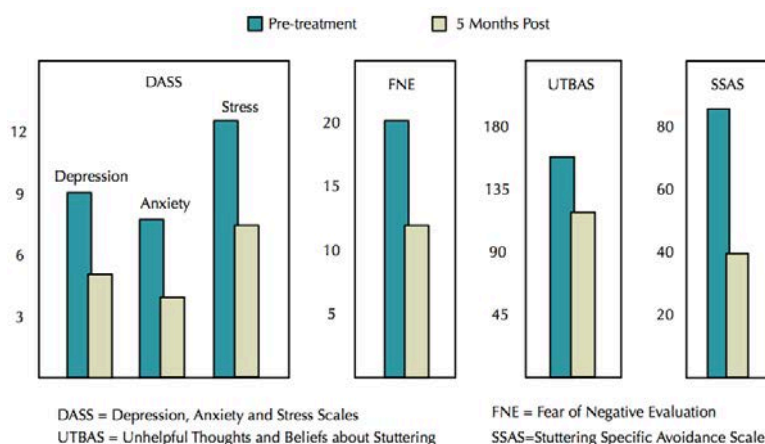
### Method

This was an international non-randomised trial with 267 participants who reported a history of stuttering and were given 5 months access to iGlebe. Participants were recruited from 23 countries, with the majority from Australia, The United Kingdom, Canada, the United States, New Zealand, and South Africa. Participants completed pre-treatment and post-treatment assessments from within the program: The Depression Anxiety and Stress Scale, the Fear of Negative Evaluation Scale, the Unhelpful Thoughts and Beliefs about Stuttering Scale, and the Stuttering Specific Avoidance Scale.

### Results

Of the 267 participants recruited, 30 did not log on, 185 did not complete Section 7 within 5 months, and three completed all sections but did not complete post-treatment assessments. Hence, the completion rate for the trial was 18.4% (49 of 267 participants) and the completion rate for iGlebe was 19.5% (52 of 267 participants). This completion rate was far superior to existing standalone Internet treatments for depression and anxiety, which attain below 7%<sup>62</sup> and around 1%.<sup>63</sup>

Without any contact from a researcher or a clinician, statistically significant pre-treatment to 5 months post-treatment reductions were reported for all measures. These results were similar to the Phase I trials of iGlebe and trials of in-clinic CBT for stuttering with a clinical psychologist. Post-treatment scores for the Depression Anxiety and Stress Scale were within typical community values. Results are shown in the figure.\*



\* Adapted and reproduced with permission: Menzies, R., et al. (2016). International Phase II clinical trial of CBTPsych: A standalone Internet social anxiety treatment for adults who stutter. *Journal of Fluency Disorders*, 48, 35–43. © 2016 Elsevier.

## A Phase II clinical trial of iBroadway with adolescents

### *Background*<sup>64</sup>

The authors of this report argued that anxiety associated with stuttering is likely to be developing during early childhood. Hence, they adapted the iGlebe program for adolescents: iBroadway.

### *Method*

Participants were 21 adolescents, ages 12–17, years who were seeking treatment for anxiety about their stuttering. Outcomes were the Diagnostic Interview Schedule for Children,<sup>65</sup> the Revised Children's Manifest Anxiety Scale,<sup>66</sup> the Children's Depression Inventory,<sup>67</sup> the Subjective Units of Distress Scale, the UTBAS scale (modified for adolescents), the OASES, parent-reported speech satisfaction, parent reported typical and worst stuttering severity, self-reported speech satisfaction and avoidance of speaking situations, and self-reported typical and worst stuttering severity. Post-treatment measures were collected 5 months after participants first accessed the program.

### *Results*

Eleven of the 21 participants completed the iBroadway modules within the allocated 5 months, which was a favourable compliance rate for internet CBT treatment. For participants who completed the modules, there was a significant post-treatment reduction of mental health diagnoses with the Diagnostic Interview Schedule for Children. There were significant post-treatment decreases for the Subjective Units of Distress Scale, the UTBAS scale, and parent reported speech satisfaction. The authors are planning continued development of the iBroadway program and further clinical trials.

## A randomised trial of iGlebe compared to in-clinic CBT

### *Background*

This trial<sup>68</sup> was designed to evaluate the relative effects of iGlebe and CBT treatment by a clinical psychologist.

### *Method*

The design was a two-arm noninferiority randomised controlled trial; a method designed to determine whether one treatment is not inferior to another. Assessments occurred at pre-randomisation and at 6 and 12 months post-randomisation. Participants were 50 adults with stuttering who were seeking anxiety treatment, 23 of whom were randomised to receive iGlebe treatment, and 27 of whom were randomised to receive in-clinic CBT by a senior clinical psychologist. The iGlebe treatment involved 5 months access to the program, and the in-clinic treatment involved from four to 11 one-hour weekly treatment sessions.

The primary outcomes were number of mood and anxiety disorders determined with a self-administered computer interview,<sup>69</sup> and the Brief FNE scale. Secondary speech outcomes were %SS based on two 10-minute, unscheduled telephone calls where participants conversed with a stranger, and a self-reported typical SR score for the previous week. Other secondary outcomes were the OASES and the Social Phobia Anxiety Inventory.<sup>70</sup>

### *Results*

Of the 23 participants randomised to receive iGlebe treatment, 18 were available for assessment at 12 months post-randomisation. Of the 27 participants randomised to receive in-clinic treatment, 24 were available for assessment at 12 months post-randomisation. Missing data at 12 months post-randomisation were accounted for by last observation carried forward (see Lecture Five). All psychological outcomes showed a consistent medium effect size with no evidence of inferiority of the iGlebe treatment on any outcome variable.

Improved self-reported SR scores at 12 months post-randomisation provided the first suggestion that CBT can improve stuttering severity. However, this result was not at all definitive, because it was not reflected in %SS scores. As the authors stated, this could have been because the %SS measure was not

valid, or because “CBT treatment produced a favourable change in the way participants perceived their stuttering severity, which prompted a posttreatment lowering of their perceived severity” (p. 10). As the authors also noted, the clinical importance of this issue warrants further research.

## A randomised trial of iGlebe supplementing speech treatment

### *Background*

The authors designed this trial<sup>71</sup> to determine whether iGlebe added to the benefits of speech restructuring treatment.

### *Method*

The design was a two-arm randomised experimental clinical trial. Assessments occurred at 6 and 12 months post-randomisation. Participants were 32 adults recruited from a stuttering treatment waiting list. Participants in both arms received Stages 1 and 2 of the Camperdown Program speech restructuring treatment in a 3-day version (see Lecture Eight). The treatment was presented without any components dealing with anxiety. In other words, the speech treatment was exclusively focused on stuttering control. The treatment included a 1-hour follow-up session each month for 5 months. Participants in the experimental arm were given access to iGlebe for 5 months after the 3-day component of the Camperdown Program treatment.

The primary outcome was %SS based on two 10-minute, unscheduled telephone calls where participants conversed with a stranger. Secondary speech outcomes were determined from typical and worst self-reported SR in eight standard speaking situations. Other secondary outcomes were avoidance of speaking situations, the OASES, UTBAS, the Brief FNE scale, the Social Phobia Anxiety Inventory,<sup>70</sup> and number of mental health diagnoses measured with a self-administered computer interview.<sup>69</sup>

### *Results*

Approximately a third of participants completed the program, and the authors noted that this was a better compliance rate than a previous trial.<sup>61</sup> At 12 months post-randomisation the groups that had access to iGlebe had significantly better results than the group that did not. This improvement occurred for typical and self-reported SR and OASES. The result was found to be robust using a statistical technique called multiple imputation to adjust for missing data due to the 18% compliance rate.

It is difficult to interpret this trial of iGlebe because, for some unknown reason, the sample was unusual for treatment-seeking adults, with only five participants having a mental health diagnosis. Improved self-reported SR scores were consistent with the finding of the randomised trial discussed previously.<sup>68</sup> This second trial of iGlebe provided another suggestion that CBT can improve stuttering severity. However, this result was not at all definitive, for the same reasons that were discussed earlier with the other trial. Clearly, this issue requires detailed exploration with a different research method to clarify it.

## Acceptance and commitment therapy

### *Background*

After the developments of Behaviour Therapy and Cognitive Behaviour Therapy, there has been a so-called “third wave” or “third generation” of cognitive behavioural therapies.<sup>72</sup> One of these is Acceptance and Commitment Therapy, commonly known as ACT. A user guide to ACT is available.<sup>73</sup> This treatment differs from CBT because it focuses on “awareness, acceptance, and understanding the context of thoughts rather than challenging and changing their content” (p. 123).<sup>74</sup> Acceptance and commitment therapy, along with several other third wave therapies, has in common with CBT that it incorporates mindfulness training,<sup>†</sup> although with a greater emphasis. A definition of mindfulness is

<sup>†</sup> The attentional training in Section Six of iGlebe described earlier is a mindfulness procedure.

“the awareness that emerges through paying attention on purpose, in the present moment, and nonjudgmentally to the unfolding of experience moment by moment” (p. 145).<sup>75</sup> The overall purpose of ACT is

to undermine the grip of the literal verbal content of cognition that occasions avoidance behavior and to construct an alternative context where behavior in alignment with one's value is more likely to occur. (p. 651)<sup>72</sup>

There have been many systematic reviews of ACT for interventions related to anxiety. For stuttering, a report found an association between a measure of mindfulness and OASES scores with stuttering participants.<sup>76</sup>

#### *Acceptance and commitment therapy with adults who stutter*

A preliminary case study explored ACT with eight adults,<sup>77</sup> followed by a speech treatment program, but reported no striking effects. There has been a description of how ACT may pertain to those who stutter,<sup>78</sup> and a subsequent description of an ACT package tailored specifically for stuttering,<sup>79</sup> and a data-based report about a similar program.<sup>80</sup> In the latter report, 20 participants received eight 2-hour group therapy sessions, with 10 participants per group. The report is difficult to interpret because participants received a combined package of speech treatment and ACT. As such, any psychological improvement could have occurred because of the speech component rather than the ACT component. And conversely, any speech improvements could have occurred because of the ACT component. The speech treatment was described as “fluency shaping activities, speech rate control, speech naturalness and self-administered timeout for stuttered moments” (p. 291).<sup>80</sup> Results at 3 months follow-up showed statistically significant improvements for stuttering severity during speech at the clinic while talking to a clinician, and improvements of OASES scores. Improvements were also shown in psychometric measures reflecting the success of the ACT therapy process. However, replications of the effects of ACT on stuttering will need to occur before it can be compared with what is known about the effects of CBT.

A report followed from the earlier description of an ACT package for stuttering,<sup>79</sup>—“fluency and Acceptance and Commitment Therapy for Stuttering”—and was described as a feasibility study.<sup>81</sup> Twenty-nine adults received the package, which involved eight 60–90-minute sessions. Some participants received individual treatment, some received group treatment, some received telehealth treatment, and some received mixed treatment formats. Assessments occurred at pre-treatment, immediately post-treatment, and at 3- and 6-months post-treatment. As with the earlier report,<sup>80</sup> improvements of all measures were reported: %SS, OASES, self-efficacy, and psychological flexibility. However, as also with the earlier reports, results are difficult to interpret because of possible interactions between the speech and psychological components of the treatment.

Another pertinent report<sup>82</sup> is difficult to interpret. Ten stuttering participants were randomly allocated to receive CBT or CBT plus mindfulness training. The report presented data for the entire 10 participants, showing improvements across a range of psychological measures. However, no data were presented to suggest that the addition of mindfulness training improved the effects of CBT. Another report<sup>83</sup> randomly allocated 36 adult stutterers to receive either (1) group CBT plus group speech restructuring, or (2) group “mindfulness and acceptance” therapy plus group speech restructuring. Results showed that the former group had superior psychological outcomes. However, a caution with interpreting these results is that it is not known whether the speech restructuring treatment contained any CBT components.

#### **Inquiry Based Stress Reduction**

A randomized controlled trial<sup>84</sup> explored an intervention known as Inquiry Based Stress Reduction. This is a meditation technique that identifies thoughts associated with stress. Then, in a meditative process, those thoughts are cognitively challenged and “reframed” into opposite thoughts. The intention is to “to experience situations that were previously perceived as stressful with peace of mind and connectedness” (p. 4).<sup>84</sup>

Participants in the trial were 65 adult stutterers, recruited from the Israeli Stuttering Association, social networks, and advertising. Twenty-eight were randomised to the treatment group and 28 to a no-treatment control group. The treatment was provided to the experimental group for 12 weekly, group sessions of 3.5 hours, with 14 participants in each group. Assessments occurred pre-treatment, at the end of treatment, and at 1-month follow-up. Assessments were the OASES-A, the State-Trait Anxiety Inventory,<sup>85</sup> the Psychological Flexibility Questionnaire,<sup>86</sup> and the Satisfaction-with-Life Scale.<sup>87</sup>

Results showed statistically significant improvements in all measures at 1 month follow-up for the experimental group compared to the control group. Significant improvements occurred for the four OASES subscales as well as the Total Score, which was 3.1 pre-treatment and 2.3 at 1 month follow-up. This represents a change from moderate-severe to moderate impairment.

Although the follow-up period was short at 1 month, these results are promising and require replication. They appear to have potential economic benefit, considering that groups of 14 participants received 14 hours of treatment, which amounts to 3 hours of treatment each.

### Self-compassion Intervention

The concept of self-compassion involves six components: “increased self-kindness, common humanity, and mindfulness as well as reduced self-judgment, isolation, and overidentification” (p. 193),<sup>88</sup> and is measured with the Self-Compassion Scale.<sup>89</sup> There are data to suggest that self-compassion is associated with anxiety.<sup>88</sup> A study<sup>90</sup> reported no differences between adult stutterers and controls on the Self-Compassion Scale. However, an association was found between self-compassion scores and OASES scores. The same researchers followed up with a pilot study of a standalone online self-compassion intervention.<sup>91</sup>

The online intervention comprised six modules: “Introduction to self-compassion, Mindfulness, Self-kindness, Common humanity, Meeting difficult emotions, Taking it forward” (p. 4). Each module required 35–45 minutes to complete, and participants completed one module each week. Fifteen adults participated and were compensated for their efforts with a gift voucher, and 10 (67%) completed the program. Post-treatment assessments were conducted immediately after completion of the final module. For those 10 participants, significant pre-post differences were reported for the Total Self-Compassion score and three of six subscales. A pre-post difference in overall OASES impact scores was reported, although both scores remained in the moderate-severe impact range. The standalone website is not currently available for public use.

## SUMMARY

Some clients who present at speech clinics with clinically significant anxiety will require intervention for it. This presents a challenge for speech-language pathologists, for whom anxiety management is typically not a primary professional domain. However, there are anxiety measurement procedures suitable for speech-language pathologists, who may wish to provide anxiety treatment if they have appropriate experience and professional preparation. There is evidence that cognitive behaviour therapy is efficacious for treating the social anxiety of those who stutter. Additionally, there is evidence that a standalone internet social anxiety treatment is efficacious. Hence, speech-language pathologists might recommend it for their clients; it has no cost and requires no clinical psychology expertise. This could prove to be a significant advance for speech-language pathologists who do not have professional qualifications for anxiety management.

Those summary comments pertain to adults and adolescents. However, they do not apply to younger clients. A systematic review of psychosocial effects of treatment for children ages 6–12 years<sup>92</sup> provided a more guarded conclusion about that age group:

Perhaps the most informative feature of the present review is not so much what emerged but what did not emerge from the review. Given the evident need for managing psychosocial impacts of stuttering during the school years, the existing literature does not reflect this contemporary perspective. Only 22 clinical reports, to date, have measured and documented any psychosocial

outcomes after treatment. Clearly, therefore, future clinical research is required to advance the evidence base for managing stuttering during the school years.  
(p. 7)



APPENDIX ONE

The UTBAS-6 scale<sup>6</sup>

**UTBAS-6**  
**Brief Version of the Unhelpful Thoughts and Beliefs About Stuttering Scales**

		Using the following scale, please read each item below and circle the number which most accurately describes you in terms of: (1) how FREQUENTLY you have these thoughts, (2) how much you BELIEVE these thoughts, (3) how ANXIOUS these thoughts makes you feel.														
		How FREQUENTLY I have these thoughts					How much I BELIEVE these thoughts					How ANXIOUS these thoughts make me feel				
		1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
1	I'll never be successful because of my stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
2	People will think I'm incompetent because I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
3	People will think I'm strange	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
4	I don't want to go – people won't like me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
5	What's the point of even trying to speak – it never comes out right	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
6	I'll never finish explaining my point – they'll misunderstand me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5

Note: items 1-6 of the UTBAS-6 are taken from the original UTBAS scales (items 15, 19, 23, 35, 50, and 60, respectively)

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## APPENDIX TWO

An example of an UTBAS-6 form completed by a client with clinically significant anxiety

Using the following scale, please read each item below and circle the number which most accurately describes you in terms of: (1) <u>how</u> FREQUENTLY you have these thoughts, (2) how much you BELIEVE these thoughts, (3) how ANXIOUS these thoughts makes you feel.																
1=never or not at all 2=rarely or a little 3=sometimes or somewhat 4=often or a lot 5=always or totally		How FREQUENTLY I have these thoughts					How much I BELIEVE these thoughts					How ANXIOUS these thoughts make me feel				
1	I'll never be successful because of my stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
2	People will think I'm incompetent because I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
3	People will think I'm strange	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
4	I don't want to go – people won't like me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
5	What's the point of even trying to speak – it never comes out right	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
6	I'll never finish explaining my point – they'll misunderstand me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5

Note: Items 1-6 of the UTBAS-6 are taken from the original UTBAS scales (items 15, 19, 23, 35, 50, and 60, respectively)

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