

**MARK ONSLOW**

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**STUTTERING  
AND ITS  
TREATMENT  
ELEVEN LECTURES**

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**APRIL  
2018**

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To Jerry Siegel  
1932–2014

## **PREFACE**

I wrote these lectures simply so that I would not have to present them verbally to students of speech-language pathology. Instead, students read them in advance and during class apply their content to professional practice.

The lectures are introductory for a student of speech-language pathology who is learning to provide health care for those who stutter. That being said, perhaps they will be of interest to a broader audience within the speech-language pathology discipline. This text is freely downloadable from the website of the Australian Stuttering Research Centre at <http://sydney.edu.au/health-sciences/asrc> and 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 appears on the cover and at the top right of alternate pages.

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. And more thanks are due to my wife Anne Skyvington than to anyone. She supported and somehow managed to tolerate me during the writing.

Apart from those broad influences, however, I alone am responsible for the content and structure of these lectures. They constitute a personal view about the course content that students of speech-language pathology need during professional preparation to provide health care for stuttering. That personal view includes judgements about the topics and research publications that students need to be aware of, and judgements about those that are beyond the scope of an introductory course.

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## CITATIONS ADDED TO THIS EDITION

The December 2017 edition has been revised to include the following publications.

- Boyle, M. P. (2018). Enacted stigma and felt stigma experienced by adults who stutter. *Journal of Communication Disorders*, doi: <https://doi.org/10.1016/j.jcomdis.2018.03.004>
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## LECTURE ONE      BASIC INFORMATION

Terms	
The disorder	1
Other terms	1
Those who have the disorder	1
When people stutter	2
When people do not stutter	3
Stuttering moments	3
A summary of recommended formal and informal stuttering terms	4
Defining stuttering	
There is no single definition of stuttering	5
Dictionary definition	5
Internal definition	6
Perceptual definition	6
Describing stuttering moments	
Taxonomies	7
Unambiguous stuttering moments	7
A taxonomy	7
Stuttering behaviours combine in one stuttering moment	10
Some practical examples of describing stuttering	11
Distribution of stuttering moments	
The influence of spoken language	12
Adaptation, consistency and adjacency	13
Identifying stuttering	
Not a clinical challenge	14
Speech and language disorder comorbidity	15
Legal stuttering identification	16
Screening for early stuttering	16
Theoretical perspectives about stuttering identification	17
Disorders to distinguish from stuttering	17
An unusual case history	19
Guidelines for interacting with those who stutter	
An important topic	19
Two caveats about eye contact	20
Conditions that reduce or eliminate stuttering	
The fluency inducing conditions	20
Verbal response contingent stimulation	21
Auditory feedback	21
Summary	

## LECTURE TWO      MORE BASIC INFORMATION

How stuttering affects people	
Speech impact	34
Quality of life impact	34
Occupational impact	35
Educational impact	35
Stuttering stereotypes	36
Guidelines Anticipation of stuttering	38
Social anxiety	39
Personality	41
Stuttering and genetics	
Background	41
Familial incidence	41
Twin studies	42
Family aggregation studies	43
Biological genetic evidence	44
A mouse model of stuttering	44
Conclusions	44
Brain structure and function	
Mounting evidence	45
Two current hypotheses	45
The critical issue	45
Clinical applications of neuroimaging research	46
Stuttering Epidemiology	
Epidemiology	46
The value of stuttering epidemiology	47
Epidemiology and public health	47
Point prevalence of stuttering	
Point prevalence	47
Two essential caveats	47
Estimates of stuttering point prevalence	48
A large data set	49
Cumulative incidence of Stuttering	
Cumulative incidence	49
Childhood cumulative incidence	49
Lifetime cumulative incidence	52
Stuttering onset	
Onset occurs during the pre-school years	52
Onset can be sudden and severe	53
Repeated movements are prominent at onset	53
More boys and men are affected than girls and women	53
Is stuttering onset predictable?	54
Natural recovery from early stuttering	
What is the natural recovery rate?	55
Two essential caveats	55
Prospective reports of natural recovery beyond the pre-school years	55
Prospective reports of natural recovery during the pre-school years	56
Is natural recovery predictable?	57
Summary	

## LECTURE THREE      THE CAUSE OF STUTTERING

Two reasons causality is clinically important	
Explaining cause to clients and parents	72
Treatment credibility and expectancy	72
An example of a clinically influential causal theory	
The Diagnosogenic Theory	72
The rise	72
The fall	73
Testing causal theory of stuttering	
Introduction	74
Testability of a theory	74
Explanatory power of a theory	74
Multifactorial models of stuttering causality	
The fundamental proposition	76
The Demands and Capacities Model	76
Other multifactorial models	77
Testability	78
Explanatory power	78
The future of multifactorial models	79
The Interhemispheric Interference Model	
The fundamental proposition	79
Testability	80
Explanatory power	80
The future of the Interhemispheric Interference Model	81
The Covert Repair Hypothesis	
The fundamental proposition	82
Testability	82
Explanatory power	83
The future of the Covert Repair Hypothesis	84
The EXPLAN Theory	
The fundamental proposition	84
Testability	85
Explanatory power	85
The future of the EXPLAN Theory	86
The P&A Model	
Background	86
The fundamental proposition	87
Testability	88
Explanatory power	88
Epilogue	
Summary	



## LECTURE FOUR      CLINICAL MEASUREMENT OF STUTTERING

Six reasons for clinical measurement	
Assessment	99
Communicating with clients	99
Stating treatment goals	99
Assessing progress toward treatment goals	99
Managing maintenance of treatment gains	100
Keeping track of daily stuttering severity changes	100
Percentage syllables stuttered (%SS)	
Overview	100
Percentage syllables stuttered scores are not normally distributed	101
Equipment for percentage syllables stuttered measurement	102
Limitations of percentage syllables stuttered	102
Severity rating (SR) scales	
Overview	103
Equal interval ordinal scales	104
Severity rating scores are not normally distributed	104
Reliability of severity ratings	105
A severity rating scoring guide	105
The clinical population as reference	106
Advantages of severity ratings	107
The relation between %SS and SR	
A strong relationship	107
Repeated movements and fixed postures	107
Percentile ranks for %SS and SR	108
The relation between %SS and SR during treatment	108
Syllables per minute (SPM)	
Speech naturalness (NAT) measurement	
Why measure speech naturalness?	108
A scale of speech naturalness	109
Stuttering-Like Disfluencies	
The Stuttering Severity Instrument (SSI-4)	
The Overall Assessment of the Speaker's Experience of Stuttering (OASES)	
The Wright and Ayre Stuttering Self-Rating Profile (WASSP)	
A simple speech satisfaction scale	
Summary	
Appendix One	

## LECTURE FIVE      EVIDENCE-BASED PRACTICE WITH STUTTERING

What is evidence-based practice?	
Speech-language pathology and evidence-based practice	
What evidence-based practice is not	
Not a rulebook	118
Not a source of all clinical knowledge	118
Not a replacement for common sense	119
How to do evidence-based practice	
Step One: Find out what the client needs	119
Step Two: Find the relevant evidence	120
Step Three: Do the treatment and evaluate its effects	121
Scientific standards for clinical evidence	
Peer-reviewed scientific journals	121
Hierarchies of evidence	121
Detailed methodological critique	122
Clinical trials of stuttering treatment	
What is a clinical trial?	122
Clinical trial standards	123
Phases of clinical trial development	
Phases I to IV	124
The CONSORT statement	124
Phase I clinical trials	124
Phase II clinical trials	125
Phase III clinical trials	125
Phase IV clinical trials	127
Finding stuttering research to inform evidence-based practice	
Finding clinical trials as they are published	128
Summary	

## LECTURE SIX      EVIDENCE-BASED EARLY STUTTERING TREATMENTS

Clinical features of early stuttering	
Early intervention with telepractice	
Telepractice	134
Advantages of telepractice early stuttering intervention	135
Three early stuttering treatments supported by clinical trials	
The Lidcombe Program	
Background	135
Overview	136
The severity rating (SR) scale	137
Percentage syllables stuttered (%SS)	138
Parent verbal contingencies	138
Some essential things about parent verbal contingencies	139
Verbal contingencies during practice sessions	140
Verbal contingencies during natural conversations	141
Stage 2	141
The Lidcombe Program problem solving	142
Clinical Strengths and limitations of the Lidcombe Program	
Strengths	142
Limitations	142
Treatments Based on Multifactorial Models: I. Palin Parent-Child Interaction Therapy	
Background	143
Overview	144
The treatment process	146
Treatments Based on Multifactorial Models: II. RESTART-DCMTreatment	
Background	147
Overview	147
Assessment	147
The treatment process	148
Clinical Strengths and Limitations of Treatments Based on Multifactorial Models	
Strengths	149
Limitations	149
The Westmead Program	
Background	149
The treatment process	150
Clinical Strengths and Limitations of the Westmead Program	
Strengths	151
Limitations	151
Summary	
Appendix One	
Appendix Two	

## LECTURE SEVEN      THE EARLY STUTTERING INTERVENTION EVIDENCE BASE

Clinical trials of one treatment	
The Lidcombe Program	157
Palin Parent-Child Interaction Therapy	159
The Westmead Program	159
Clinical trials comparing two treatments	
Lidcombe Program compared to RESTART-DCM Treatment	160
Translational research	
The Lidcombe Program	162
Randomised clinical experiments	
The Lidcombe Program	163
Data-based case studies	
The Lidcombe Program	164
A family-focused treatment approach based on a multifactorial model	165
Treatment fidelity research	
The Lidcombe Program	165
Treatment mechanisms	
The Lidcombe Program	167
Treatments based on multifactorial models	168
Treatment safety	
The Lidcombe Program	169
Treatments based on multifactorial models	169
How long does treatment take?	
The Lidcombe Program	169
How does a treatment delay affect the treatment process?	
The Lidcombe Program	171
Do case variables affect the treatment process?	
The Lidcombe Program	172
Parent experiences	
The Lidcombe Program	172
The early stuttering intervention evidence base: Summary and conclusions	
The Lidcombe Program	174
Treatments based on multifactorial models	175
The Westmead Program	175

## LECTURE EIGHT      EVIDENCE-BASED ADULT SPEECH TREATMENTS

Speech restructuring treatment	
Background	182
A brief history	183
Programmed instruction	
A technique for behavioural control	184
Performance contingent progression	184
A fundamental assumption	184
Models of programmed instruction	184
An example of programmed instruction	184
Instatement and transfer	186
The clinical trial evidence for speech restructuring treatment	
Numbers of trials	186
Effect size	186
Speech naturalness	187
Speech restructuring I: The Camperdown Program	
Background	188
Stage I: Teaching treatment components	189
Stage II: Establishing stutter-free speech	189
Stage III: Generalisation	191
Stage IV: Maintenance of treatment gains	191
Clinical trial evidence for the Camperdown Program	191
Speech restructuring II: The Comprehensive Stuttering Program	
Overview	193
Clinical trial evidence for the Comprehensive Stuttering Program	193
Speech restructuring III: Video self-modelling as a supplement	
The procedure	194
Basic research	194
A data-based case study of video self-modelling: Relapse management	194
A Phase III trial of video self-modelling: Speech restructuring supplement	195
A clinical experiment	196
A verbal response contingent treatment: Self-imposed time-out	
The procedure	197
Clinical advantages	197
Clinical trial evidence for self-imposed time-out	197
Machine aided treatments	
Background	198
Altered auditory feedback	199
Modifying phonation intervals	200
Transcranial direct current stimulation	202
Pharmacological Treatments	
Summary	

**LECTURE NINE                    EVIDENCE-BASED ADOLESCENT  
AND SCHOOL-AGE SPEECH TREATMENTS**

Speech restructuring I: Intensive smooth speech	
A Phase II trial	210
Results	210
Anxiety reduction	211
Follow-up	211
Speech restructuring II: The Comprehensive Stuttering Program	
A Phase II trial	211
Results	212
Speech restructuring III: The Camperdown Program	
In-clinic	212
Telepractice	213
Speech restructuring IV: Video self-modelling as a supplement	
A Phase III trial	214
Results	214
Verbal response contingent stimulation	
The Lidcombe Program	215
Gradual Increase in Length and Complexity of Utterance (GILCU)	215
Self-imposed time-out	216
Clinician-imposed time-out	216
Syllable-timed speech	
Method	217
A Phase I trial	217
Results	217
A hybrid treatment: Syllable-timed speech and verbal response contingent stimulation	
Method	218
A Phase II trial	218
Results	218
Machine aided treatments	
Electromyographic (EMG) biofeedback	219
Altered Auditory Feedback	220
Conclusions about the adolescent and school-age evidence base	
Speech restructuring	220
Verbal response contingent stimulation	221
Syllable-timed speech	221
Machine-aided treatments	222
Clinical Notes: Adolescents	
A life transition	222
Parents during treatment	222
Telepractice and adolescents	222
Clinical Notes: School-age children	
A period of changing tractability	223
Adaptation of the Lidcombe Program for school-age children	224
Teachers and school-age children who stutter	224
Summary	

## LECTURE TEN      STUTTERING, SOCIAL ANXIETY, AND MENTAL HEALTH

Background	
A changing view about stuttering and anxiety	231
Anxiety	
Expecting harm	231
Three components	231
Stuttering, anxiety, and anxiety disorders	
Stuttering and anxiety	232
Stuttering and social anxiety disorder	233
Stuttering and other anxiety related disorders	234
The Clark And Wells model of social anxiety disorder	
The puzzle	235
The Clark and Wells model	235
Three assumptions of the model	235
Negative self processing in social situations	235
Safety behaviours	237
Somatic and cognitive anxiety symptoms	239
Before the feared situation	239
After the feared situation	240
Anxiety impairs speech treatment	
The problem of post-treatment relapse	240
Anxiety and post-treatment relapse	240
The origins of social anxiety with stuttering: The pre-school years	
Direct evidence: Psychometrics	241
Direct evidence: Early childhood temperament	242
Indirect evidence	246
Conclusions	247
The origins of social anxiety with stuttering: The school-age years and adolescence	
Direct evidence	248
Indirect evidence	250
Conclusions	252
Stuttering, mental health, and the timing of early intervention	
Summary	

## LECTURE ELEVEN      TREATMENT OF SOCIAL ANXIETY

Speech-language pathologists and anxiety treatment

Anxiety measurement for speech-language pathologists

Background	266
The Unhelpful Thoughts and Beliefs About Stuttering (UTBAS) scales	267
The Fear of Negative Evaluation (FNE) scale	269
Subjective Units of Distress Scale (SUDS)	270
The Spence Children's Anxiety Scale	270
The Preschool Anxiety Scale Revised	271

Evidence-based anxiety treatment for stuttering

Cognitive Behaviour Therapy (CBT)	272
CBT for stuttering: A clinical randomised controlled trial	272
Development of standalone Internet CBT for stuttering	273
Program design	274
Phase I clinical trials of CBTpsych	280
A Phase II clinical trial of CBTpsych	281
Acceptance and commitment therapy	282

Summary

Appendix One

Appendix Two



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## LECTURE ONE

### BASIC INFORMATION

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#### TERMS

##### The disorder

###### *Stuttering and stammering*

Worldwide, the term *stuttering* is used most commonly to refer to this speech disorder. The term *stammering* is often used in the United Kingdom and Ireland. Most publications about the disorder, however, use the term *stuttering*.

###### *Potential confusion*

According to the American Speech-Language-Hearing Association<sup>1</sup> the disorder “is plagued with inconsistent, confusing terminology. This problem has cultural, historical, linguistic, and practical origins.” So, the following material is presented with the intention that clinical terminology for the disorder is as clear as possible.

##### Other terms

These terms sometimes are used to refer to stuttering: *dysfluency*, *disfluency*, and *nonfluency*. However, as discussed shortly, there are arguments for not using them.

##### Those who have the disorder

###### *Direct and person-first terms*

Historically, someone who has the disorder was referred to directly as a *stutterer*, and those with the disorder as *stutterers*. Person-first terminology is a different and more recent approach, intended to avoid any negative connotation from labelling someone with a disorder. Instead, reference is to someone who has a disorder. Accordingly, preferred terms would be *a person who stutters*, *someone who stutters*, or *those who stutter*.

###### *Making a choice*

When making a choice about how to refer to those who have the disorder, clinicians may be influenced by the views of clients. Some clients might prefer direct terms and some might prefer person-first terms. A useful rule of thumb is to err on the side of caution, and to use person first terms if there is any uncertainty. When writing a formal report about clients, clinicians may prefer person-first terms. Some scientific speech-language pathology journals require the use of person-first terms for stuttering.

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

For all its potential benefits, there are potential limitations with person-first terms for stuttering. Two research publications<sup>2,3</sup> raise doubt about whether person-first terminology for stuttering alters negative perceptions about the disorder. It is also the case that person-first terms invoke present tense, and this can cause awkward expression when writing with past tense. For example, the tense in neither of these wordings is completely satisfactory: “the research participants were people who stutter” and “the research participants were people who stuttered.” At least one publication<sup>4</sup> has declined to use person-first terminology in order to avoid wording problems that it causes.

Additionally, it might be argued that the semantics of person-first terminology is misleading about the nature of the disorder. It might imply that stuttering is something that speakers do when speaking, rather than something that happens to them when they speak. The latter is what in fact happens, as discussed during Lecture Three. Person-first terminology also suggests that those affected by the

disorder, and who suffer negative effects from it, will necessarily “stutter” in an obvious way. Subsequent lectures show that to be not all true.

## When people stutter

### *Stuttering and stuttering behaviour*

As well as being a term to refer to the disorder, *stuttering* can be used to refer to someone’s speech being affected by it. For example, “she was stuttering a lot yesterday,” and “stuttering on the telephone is a problem for him.”

Sometimes clinicians use the term *stuttering behaviour* (spelled *behavior* in the United States) in formal contexts such as written reports and conference presentations. The term *behaviour* to describe stuttering is a little different from the everyday use of the term. Speech *behaviour* is something that you can see or hear. An example of something that is not *behaviour* is anger. Anger can’t be seen because it is emotion, not *behaviour*. The only way to know that people are angry is for them to tell you, or to infer that they are angry from their *behaviour*.

Researchers sometimes use the term *stuttering behaviour* in scientific publications. For example, “the observers were instructed to push a button for every stuttering behaviour,” and “the stuttering behaviours reportedly began suddenly.” However, clinicians may not wish to use such formal terms when talking to clients and parents or when writing notes in client files.

It is possible for researchers to use sophisticated instruments to measure stuttering behaviour. For example, kinematic (movement) measures of lip variability during speech have been shown to distinguish children<sup>5</sup> and adults who stutter from those who don’t.<sup>6</sup> However, clinicians use more convenient verbal descriptions of stuttering behavior, as described shortly.

### *Dysfluency and dysfluent*

*Dysfluency* and its adjective *dysfluent* are often used to describe when people stutter. For example, “his speech has been dysfluent for the past week,” and “dysfluency in the workplace is a problem for her.” But, strictly speaking, there is a problem there because the opposite of those terms, *fluency* and *fluent*, as they are commonly used in English, do not specifically refer to anything about the disorder of stuttering. They refer to a range of things about the flow of speech, not just stuttering,<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.

Another problem is that *dysfluency* and *dysfluent* are used sometimes to refer to the effects of other speech or language disorders where the flow of speech is disrupted, such as dysarthria and aphasia. So the use of those terms for stuttering is not particularly precise.

The 2013 edition of the Diagnostic and Statistical Manual of Mental Disorders,<sup>8</sup> generally known as the DSM-5, introduced the term *childhood-onset fluency disorder* and presents it interchangeably throughout with *stuttering*. Arguably, this is not at all helpful, but so far seems not to have influenced the field of speech-language pathology.

### *Disfluency and disfluent*

The situation potentially is more confusing when the terms *disfluency* and *disfluent* are used to refer to stuttering. The problem is that the prefix *dis-* does not necessarily mean that something is disordered. The prefix *dys-* does mean that, but the prefix *dis-* can mean something more like different, or not usual. So *disfluency* is not an ideal term to use for stuttering. This is particularly the case when the term is used to refer to speech associated with other speech disorders, a common example being autism.<sup>9,10,11,12</sup> Lecture Four touches on a particularly confusing use of this prefix with the term *Stuttering-Like Disfluencies*.

*Nonfluency and nonfluent*

The same problem pertains to the terms *nonfluency* and *nonfluent* that are sometimes used for stuttering. These are potentially confusing because, again, the prefix *non-* does not necessarily mean disordered.

**When people do not stutter***No stuttering*

*No stuttering* and its variations are simple and non-confusing ways to describe someone's speech that has no stuttering. For example, "he reported no stuttering all last week," and "I have not heard you stutter for the past 10 minutes."

*Stutter-free speech*

The expression *stutter-free speech* is a more formal way of referring to speech that does not contain stuttering. For example, "his speech was stutter-free during a 5-minute telephone call," and "she was stutter-free during a presentation at work." Clinicians sometimes write *stutter-free speech* in formal contexts such as reports about clients and in professional or scientific publications, but they may be reluctant to use such a formal expression when speaking with clients or their parents. That issue aside, *stutter-free speech* certainly is a non-confusing term.

*Fluency and fluent*

*Fluency* and its adjective *fluent* are other ways to refer to speech that does not contain stuttering. For example, "you were fluent just then when we were talking," and "he has been fluent for weeks now." Pedantically speaking, there is the issue previously described that such terms are not stuttering specific. However, clinicians use them commonly and there is rarely any confusion when they are used to mean speech that does not contain stuttering.

*Normal disfluency and normally disfluent*

As discussed earlier, the terms *disfluency* and *disfluent* are not correct terms for when people stutter, because the prefix *dis-* does not necessarily refer to disordered speech. However, the terms *normal disfluency* and *normally disfluent* can be used correctly to refer to the usual hesitations and repetitions that can be a part of everyday speech.<sup>7</sup> Examples of normal disfluency might be "well, um, ... gosh, I don't know," and "er, I think, perhaps, um, I will have to get back to you about that."

It is necessary to refer to such normal speech events in a way that distinguishes them from stuttering. That is because those who stutter will have *normal disfluencies* in addition to stuttering. It is particularly important to get this terminology right when treating young children for stuttering. That is because when stuttering has been successfully treated and stops being a problem—which is what should occur, as discussed during Lecture Seven—it is common for parents to be overly vigilant and mistake normal disfluencies for stuttering.

Here are some examples of clinical file notes that illustrate this point: "His mother was concerned that stuttering had returned, but on clinical inspection it was obvious that she was concerned about normal disfluency," and "I made it clear to his father that John is normally disfluent sometimes and not to confuse it with the return of stuttering."

**Stuttering moments***The idea of stuttering moments*

The stuttering moment is a useful concept for clinical practice. The idea is that those affected have speech that appears to be just like anyone else except for short periods—moments—when stuttering occurs. The first documented evidence of this idea appears to have been during the early 20<sup>th</sup> Century at the University of Iowa.<sup>13</sup> The idea appeared regularly in subsequent research literature, however the first formal statement of it appears to have occurred some 30 years later:<sup>14</sup>

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 stutters, by regarding it crucially not as a more or less constant condition, but as intermittent responses. (p. 13)

A series of experiments which concluded during the late 1980s<sup>15,16,17,18,19,20</sup> established, overall, that the speech of those who stutter sounds normal apart from stuttering moments. So, in a clinical sense, it is appropriate to think of stuttering as momentary speech disturbances surrounded by otherwise normal sounding speech. In reality, though, it is possible that the speech physiology of those who stutter is unusual whenever they speak, but the only perceptible problems are what observers label as stuttering moments.

#### *Stutters, stutters, dysfluencies, disfluencies*

The idea of stuttering moments has been popular since its inception, and to this day clinicians use it during clinical practice. In formal reports they may write *moment of stuttering*, *stuttering moments*, or *stutters*, but those terms are generally thought to be too formal for speaking with clients and parents. It is more common for clinicians to refer to *stutters* or a *stutter* during clinical practice. They may also use terms discussed previously—along with their potential limitations—to describe moments of stuttering: *dysfluencies*, *disfluencies*, or *nonfluencies*.

#### *Stuck words, bumpy words*

When talking to young children about their stuttering, clinicians need a different kind of language. Popular terms with young children are *bumpy word* or *bumpy words*, and sometimes, *stuck word* or *stuck words* are used. The important thing to remember here is the need to communicate effectively with the child about stuttering, so any terms that do that are useful.

### **A summary of recommended formal and informal stuttering terms**

	RECOMMENDED	NOT RECOMMENDED	
THE DISORDER	<i>stuttering</i>	<i>dysfluency</i> [2]	
	<i>stammering</i> [1]	<i>disfluency</i> [2]	
		<i>nonfluency</i> [2]	
THOSE WHO HAVE THE DISORDER	<i>person who stutters</i> [3]	<i>stutterer</i> [4]	[1] May be preferable in the United Kingdom and Ireland
	<i>someone who stutters</i> [3]		
	<i>those who stutter</i> [3]		
WHEN PEOPLE STUTTER	<i>stutters</i>	<i>dysfluency</i> [2]	[2] May not be clear that you are referring to stuttering
	<i>stuttering</i>	<i>dysfluent</i> [2]	[3] Person-first terms are a conservative option
	<i>stuttering behaviour</i> [5]	<i>disfluency</i> [2]	[4] Many prefer person-first terminology
		<i>disfluent</i> [2]	
		<i>nonfluency</i> [2]	[5] For use in formal contexts
WHEN PEOPLE DO NOT STUTTER	<i>no stuttering</i>	<i>fluency</i> [2]	[6] For use with young children
	<i>stutter-free speech</i> [5]	<i>fluent</i> [2]	
STUTTERING MOMENTS	<i>stutters</i>	<i>dysfluencies</i> [2]	
	<i>stuck words</i> [6]	<i>disfluencies</i> [2]	
	<i>bumpy words</i> [6]	<i>nonfluencies</i> [2]	

## 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 definition would contain words to make it clear who does and who does not have the disorder. Unfortunately, though, after a vigorous debate for a decade beginning during the early 1980s, the search for such a workable and generally agreed stuttering definition ground to a halt without resolution.<sup>21,22,23,24,25,26,27,28,29,30,31,32,33</sup>

That debate established three approaches to defining stuttering. An important point here is that none of the three definitions can be considered completely satisfactory.<sup>30</sup> They all have limitations but also some strengths that make them useful in some professional contexts. Each of them is useful in different ways.

### Dictionary definition

#### *The World Health Organisation definition*

The most common definitions of stuttering are known as dictionary definitions.<sup>30</sup> They are also known as symptomatic definitions and objective definitions. The World Health Organisation classifies stuttering as a disability, and in 1977 offered what seems to be the most popular definition to date:<sup>34</sup>

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)

A more recent World Health Organisation definition<sup>35</sup> has so far attracted less attention.

#### *Wingate's definition*

Another older and commonly cited dictionary definition of stuttering is Wingate's.<sup>36</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)

#### *Limitations of dictionary definitions*

These and other dictionary 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. That is because there are no observable speech events that can be recorded with words and which categorically distinguish between stuttering and normal speech.<sup>37</sup> At some time everyone has normal 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. That definition has been criticised also because it contains "qualifiers and imprecise terms" (p. 17),<sup>38</sup> such as "readily,"

"sometimes" and "usually," and because speech dimensions such as "controllable" and "involuntary" are not observable,<sup>39</sup> as should be the case with an objective dictionary definition.

Another problem with dictionary definitions is that they overlook a well-known group who are never or rarely heard to stutter but who are troubled by the disorder nonetheless. This is commonly referred to as covert stuttering.<sup>40</sup> For such people, speech behaviours cannot be used to define their disorder because they rarely, or sometimes never, show any observable stuttering moments.

#### *A strength of dictionary definitions*

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

### **Internal definition**

#### *Perkins' definition*

Perkins' definition<sup>24,26</sup> is stuttering as "temporary overt or covert loss of control of the ability to move forward fluently in the execution of linguistically formulated speech" (p. 431).<sup>26</sup> This is referred to as an internal definition because "loss of control" refers to a speaker's experience. This contrasts it with the intended objective, observable features of dictionary definitions.

#### *Limitations of internal definition*

It has been argued that internal definition is more a statement about the nature of the disorder than a definition.<sup>41</sup> Also, this definition has in common with dictionary 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 problem is that clinicians cannot observe "loss of control" because it is a private event, not a behaviour. Of itself, that is not a particularly serious problem, but overall it is better if clinicians can observe the presence of the disorder.

#### *A strength of internal definition*

The internal definition of stuttering certainly is a valid one, because stuttering is fundamentally a private experience for those affected. The proponents of this definition even conducted an experiment purporting to verify this.<sup>42</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.

Clinicians rely on internal definition of stuttering during routine clinical measurement of stuttering severity. As discussed during 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.

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 the disorder, and a positive shift of how they feel it affects them.

### **Perceptual definition**

#### *Bloodstein's definition*

Bloodstein's definition<sup>29</sup> is that stuttering is "whatever is perceived as stuttering by a reliable observer who has relatively good agreement with others" (p. 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.

*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>39</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. However, there are imposing practical aspects of such mentoring. And there is a risk that different clinical communities, such as those in different countries, may develop different perceptions about what stuttering is and what it is not. There is some evidence that this may occur.<sup>43</sup>

*Strengths of perceptual definition*

An advantage of perceptual definition is that it is procedurally simple and clinically workable if the required consensus exists. 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 such parents are reliable observers who have "relatively good agreement with others," and so they are using a perceptual definition of stuttering.

**DESCRIBING STUTTERING MOMENTS****Taxonomies**

Johnson developed the first system for classification of stuttering moments.<sup>44</sup> That 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 that initial taxonomy.<sup>45,46</sup> The better-known terms added to Johnson's original taxonomy are *disrhythmic phonation, block, blockage, and tense pause*. All those taxonomies deal with stuttering during early childhood, with the exception of one.<sup>47</sup> Presumably, this is because of the historical influence of two theoretical perspectives about early stuttering, which are overviewed shortly: the Diagnosogenic Theory and the Continuity Hypothesis.

**Unambiguous stuttering moments**

For the most part, those who come to a clinic complaining that they or their children stutter will be referring to many unambiguous stuttering moments that occur during each day. The term unambiguous stuttering moments refers to moments during speech that, to an observer, are clearly stuttering and not normal disfluency.

This does not mean that a clinician will never be undecided about whether a particular speech event is a stuttering moment or a normal disfluency. To the contrary, that is certain to occur, particularly with young children. However, during clinical practice this is not normally an issue. An exception is the situation described previously after successful treatment of young children, when parents may need guidance with being sure of the distinction between a stuttering moment and a normal disfluency.

**A taxonomy***Overview*

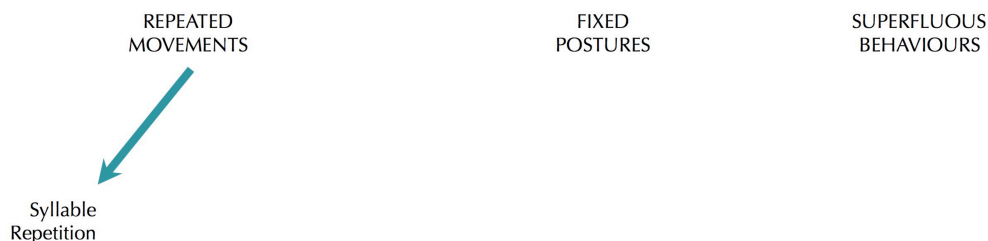
The following method to describe unambiguous stuttering moments arguably has some advantages.<sup>45</sup> It was developed for use with stuttering clients of all ages and it describes speech behaviours only; it contains no reference to anything that cannot be observed. Additionally, it appears that with some clinical experience it can be used reliably.<sup>48</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>36</sup>

*Repeated movements*

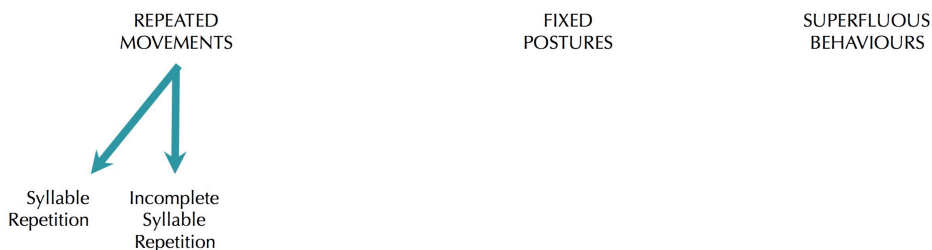
If an unambiguous stuttering moment contains a recurring movement, it is a *repeated movement*. Commonly, clinicians refer to these as repetitions. There are three different types of repeated movements.

The first type of repeated movements is *syllable repetition*.



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.

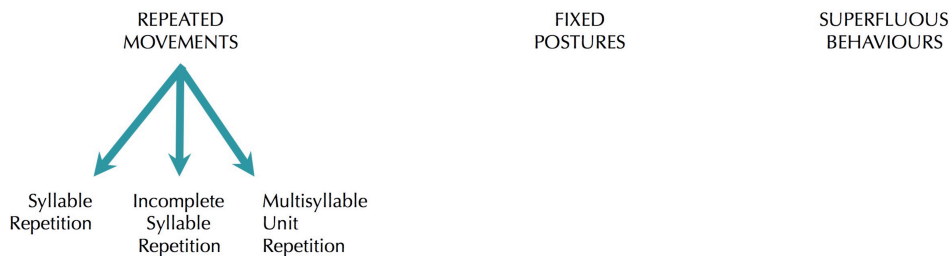


Some of the distinctions between a syllable repetition and an incomplete syllable repetition are quite obvious. For example “can-can-can-can” might be heard as a repetition of the entire syllable, with all its phonemes. But with “ca-ca-ca-ca-can” the speaker has produced only the first two phonemes of the syllable before eventually getting it right. In which case it is an incomplete syllable repetition. Careful listening to stuttering moments may be needed to make that distinction.

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 “l-l-l-l-l.” At first it might seem that this could be nothing but a syllable repetition, but again, careful listening is needed. The word “l” is a diphthong in most spoken English and the speaker might not complete the two vowel-like parts of this, and instead something like “uh-uh-uh-uh” might be heard while attempting to say “l.” 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.



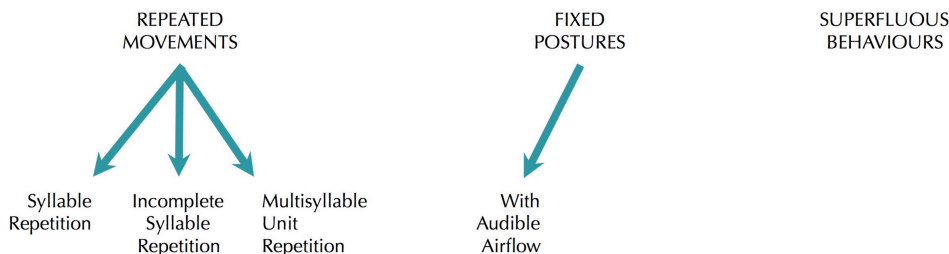


Examples of multisyllable unit repetition would be “I was-I was-I was-I was hoping,” “I think that-I think that-I think that-I think that,” and “then-I then-I then-I.”

*Fixed postures*

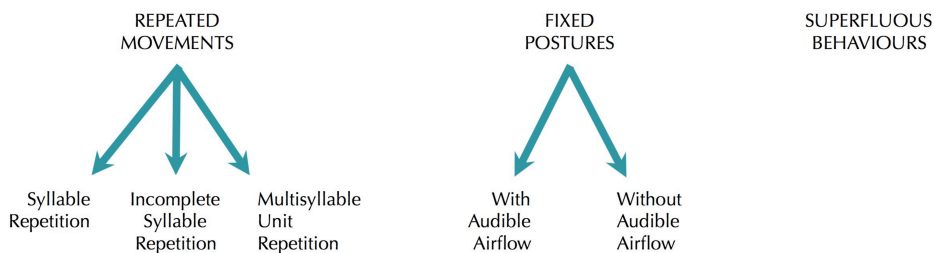
*Fixed postures* are in a sense the opposite kind of stuttering behaviour to repeated movements because they are not an abnormality of movement but an abnormality of no movement. During fixed postures what normally is seen to move during speech—mostly mouth, jaw and lips—stops moving. It can stop moving for a period so short that it might be necessary, when learning to describe stuttering moments, to look at a video carefully many times to detect it. It is far more obvious when fixed postures happen for quite 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*.

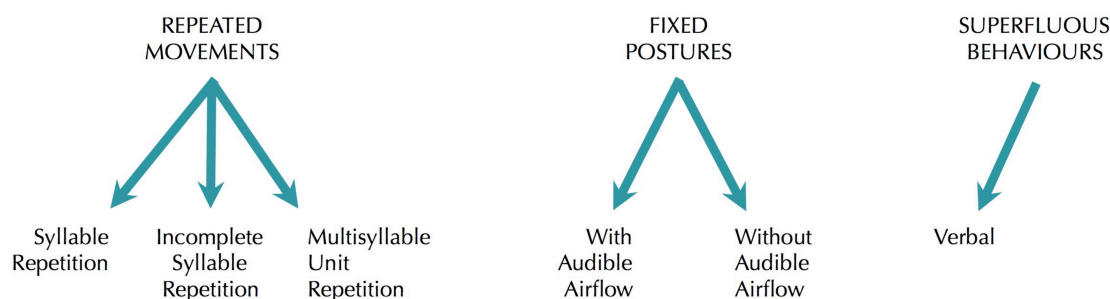


During these no airflow is audible. But it is necessary to listen carefully to be sure that there really is no sound. Sometimes the audible airflow during fixed postures can be barely audible. Clinicians often refer to fixed postures without audible airflow as “blocks,” because they give the impression that something is blocking speech.

### *Superfluous behaviours*

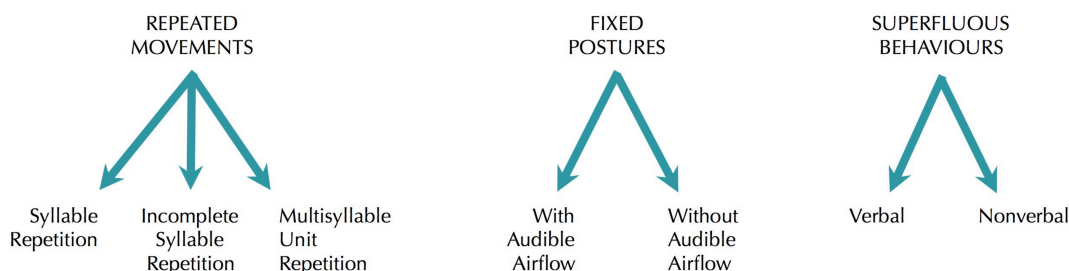
The final category of stuttering moments is *superfluous behaviours*. They are redundant to the intended meaning of the utterance as it normally would be spoken, hence the term superfluous. These are often the most disfiguring of the observable problem behaviours of stuttering.

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>44</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 aberrant fluctuations in pitch and loudness. Stuttering is an idiosyncratic disorder. It is rare to see two people whose stuttering looks identical, and nonverbal superfluous behaviours are the most idiosyncratic features of the disorder.

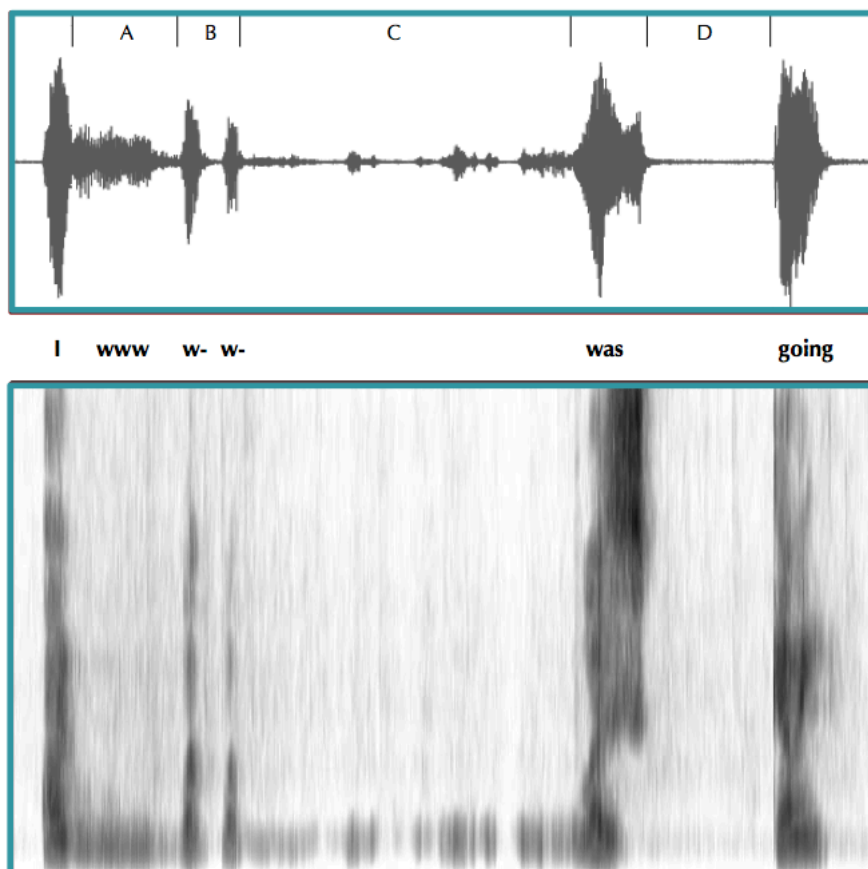
### **Stuttering behaviours combine in one stuttering moment**

The seven stuttering behaviours described with this taxonomy, or with any taxonomy, are not mutually exclusive.<sup>37</sup> One, many, or even all of the seven stuttering behaviours, can be present during one

stuttering moment.<sup>49</sup> In fact, it is rare for a stuttering behaviour to have only one of the seven stuttering behaviours. It seems that the distribution of stuttering behaviours is the same in Cantonese as it is in English.<sup>50</sup> Yet Cantonese differs markedly from English because it is tonal and syllable-timed.

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-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-can-can-can I-can I.”

The waveform (top panel) and spectrogram (bottom panel) in the 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).



### Some practical examples of describing stuttering

#### *Communicating with other clinicians*

When writing about a client to another clinician who is expert with stuttering, it may be preferable to incorporate formal terminology of the kind just described:

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, typically 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 and to the left. None of Mr Williams' stuttering moments involved repeated movements.

## 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>51</sup> showed that their occurrence is lawful to a considerable extent. Stuttering was shown to occur more commonly on consonants than vowels, with the vast majority of stuttering—more than 90%—occurring at the initial sounds of words. That finding has been replicated<sup>52,53,54,55,56,57,58,59,60</sup> many times, although failure to replicate has occurred.<sup>61</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>52,54,55,60,62</sup> although, again, a failure to replicate has occurred.<sup>63</sup> The effect has been reported also for the first word of clauses.<sup>64</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>65,66,67,68,69,70,71</sup> However, queries have been raised that such repeated movements may not be connected with stuttering.<sup>70,71,72</sup> One report<sup>71</sup> found other dysfluency types than repeated movements to occur at the end of words, but reported that they are difficult to identify perceptually.

#### *Difficult sounds*

Clients commonly find certain sounds particularly troublesome because stuttering is likely to occur with them. The seminal report mentioned previously<sup>51</sup> showed individuality among those who stutter for sounds that are stuttered more often, and hence considered to be "difficult." Another report from the same laboratory<sup>73</sup> verified that finding, by showing that there is no general rule for those who stutter about which sounds are difficult. There are reports that more stuttering is likely to occur with a second language than a first.<sup>74,75</sup>

#### *Content words*

Another early report from the University of Iowa<sup>76</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 on content words than function words. This finding has been replicated many times,<sup>53,60,61,62,77,78,79,80,81,82,83</sup> and there is some suggestion that the effect may be language specific, with a report that it does not occur in Arabic.<sup>84</sup> However, it has

<sup>†</sup> It is a general rule that research findings are not particularly believable unless they have been reported by researchers who are completely independent of the researchers who found them originally.

been reported in Persian.<sup>85</sup> One report<sup>86</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 this situation is reversed. There are reports of more stuttering on function than content words for English,<sup>87,88,89,90</sup> German,<sup>91,92</sup> Spanish,<sup>93</sup> and Spanish-English bilingual<sup>94</sup> children. Two reports<sup>80,82</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 discussed during Lecture Three.

### *Stressed syllables*

Some reports have found that stressed syllables are stuttered more often than unstressed syllables,<sup>52,95,96,97,98,99</sup> although others have failed to find such an effect.<sup>58,59,100,101</sup> A report has extended such research to lexical tone with 20 Mandarin speaking Taiwanese pre-schoolers, with a mean age 4 years 9 months.<sup>102</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. 1).<sup>102</sup> That 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>103</sup> found no differences for stuttering moments across its six tones.

### *Grammatical complexity*

There have been findings of increased stuttering with increased utterance length, which is associated with increased syntactic complexity.<sup>58,104,105,63</sup> Those findings have been replicated with children,<sup>106,107,108,109,110,111,112,113,114,115</sup> although it is probably fair to say that the findings are not as marked and consistent as with adults. Consistent with those findings are reports that long words are stuttered more often than short words.<sup>116,117</sup>

### *Clustering*

Another feature of stuttering is clustering, which is the occurrence of a series of stuttering moments at one time during speech. This has been reported several times for early stuttering during the pre-school years,<sup>118,119,120</sup> and also with adults.<sup>121,64</sup>

### *How lawful?*

An early report<sup>81</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>116</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 lawfully, but it is not possible to fully account for the occurrence of every stuttering moment.

## **Adaptation, consistency and adjacency**

### *A mysterious 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 the disorder during the last century.

Why the adaptation effect occurs is a mystery. There are data to suggest that it occurs because of subtle changes to speech motor function that occur over successive readings,<sup>122</sup> and there are data to suggest the opposite.<sup>123</sup> There is some evidence that motor learning may explain it.<sup>124</sup> That explanation is bolstered by evidence that the adaptation effect seems to occur with the “stuttering-like disfluencies” (see Lecture Four) of Parkinson disease.<sup>125</sup> The effect might also be explained if anxiety about speaking systematically reduces after several readings.<sup>126,127</sup>

*Even more mysterious ...*

Making the adaptation effect even more puzzling is the consistency effect and the adjacency effect. The consistency effect is that stuttering tends to occur on the same words during repeated readings of a passage, suggesting anxiety about specific words.<sup>128,129</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>130,131</sup> Both these effects might be explained in terms of anxiety about certain words.

## IDENTIFYING STUTTERING

### Not a clinical challenge

#### *Chronic stuttering*

Generally, clinicians don't need to diagnose stuttering in a clinic and tell people that they have the disorder. Those who have stuttered for much of their lives will be fully aware of it. So, those with chronic stuttering who present to clinics seeking clinical help will nearly always be correct that they stutter. The only clinical task is to confirm the presence of stuttering rather than some other disorder, as discussed shortly. The identifiable nature of the disorder with adults and adolescents seems to apply even to other languages. According to a recent conclusion from three papers dealing with the topic,<sup>132,133,134</sup> there is "little difference in judges' abilities to identify stuttering in other languages" (p. 222).<sup>135</sup>

#### *Pre-school children*

During the 1980s and 1990s many protocols were developed for distinguishing between stuttering and normal disfluency with pre-school children.<sup>136,137,138,139,140,141,142,143,144,145,146</sup> This topic was considered so important that a prominent clinical journal published two reviews of the area during the early 1990s.<sup>147,148</sup> However, during this century there have been no further empirical developments or reviews published about the topic, which might reflect that such protocols are currently thought to be clinically unnecessary.<sup>152</sup> Some authorities in the field, after an earlier attempt to develop a differential diagnostic protocol,<sup>142</sup> have endorsed such an opinion.<sup>154</sup>

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)

A report published shortly after that statement confirmed it. Pre-schoolers who stuttered and a control group were studied speaking with parents and clinicians at home and at the clinic, with the conclusion that<sup>149</sup>

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)

Consistent findings, attesting to the ease of stuttering identification during the pre-school years, emerged from a study of nine 3–5 year old Icelandic speaking children.<sup>150</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. However, an earlier study of pre-schoolers who stuttered presented conflicting findings:<sup>151</sup>

situational variability can make it more difficult for clinicians to correctly identify and document a child's need for treatment based upon objective measures of the child's speech fluency collected in a single speaking situation. (p. 199)

*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:<sup>152</sup>

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)

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>153,154</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>153</sup> Another authority stated “typically, parents of young children who stutter correctly diagnose the problem, making the professional evaluation a task of describing and quantifying the disorder rather than differentiating it from other disorders” (p. 313).<sup>154</sup>

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.

**Speech and language disorder comorbidity***An ambiguous literature*

Some research has reported how many children have stuttering and another speech or language disorder, or have stuttering comorbidities, to use the correct term. There is no doubt this will occur sometimes.<sup>155,156,157,158</sup> A study of clinicians<sup>159</sup> indicated that 44% of 467 stuttering school-age children reportedly also had a language or phonological disorder. Another<sup>160</sup> reported 34% had articulation disorders and 14% had phonological disorders. However, another report<sup>161</sup> found no such difference between stuttering and control children. A recent report<sup>162</sup> studied 58 stuttering pre-school 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>163</sup> concluded that research about the topic is ambiguous. Given such ambiguity in the literature, it is not surprising that clinicians are uncertain about concurrent management of children with comorbid stuttering and speech sound disorder.<sup>164</sup>

The same ambiguity pertains to findings about language problems with stuttering pre-schoolers and school-age<sup>†</sup> children. Many reports have found that children who stutter have language less advanced than peers<sup>165,166,167,168,169,170,171,172,173,174</sup> and many have found that not to be the case.<sup>175,176,177,178,179,180,181</sup> **Error! Bookmark not defined.** The recent 4–5 year study referred to previously<sup>162</sup> found that the children who stuttered “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>172</sup> was a meta-analysis<sup>†</sup> 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>180,182</sup> reported that available research did not support any such conclusion.

<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.

<sup>†</sup> Meta-analysis is a systematic review that synthesises evidence from numerous empirical reports.

*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 the disorder in general.<sup>158</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>175</sup>

*Identifying stuttering during other assessments*

There are some occasions when clinicians might need to identify children who are stuttering but are referred to the clinic because of other speech or language disorders. It certainly is possible, but probably rare, for parents to complain of other speech and language disorders and be unaware that a child is stuttering. It is more likely that this would occur with referrals from doctors or staff at pre-school day care centres or kindergartens.

**Legal stuttering identification**

Clinicians may be asked to provide a written report to a lawyer, or give verbal evidence in a court. 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>183</sup> Publications are available to assist clinicians with preparing such legal assessments.<sup>183,184,185</sup>

**Screening for early stuttering***The benefits of early 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 the development of the disorder, which is during the pre-school years, as discussed during later lectures.

*Screening sensitivity and specificity*

Screening is not assessment. It is typically a much briefer procedure than assessment, and 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 at screening to be stuttering according to the results of a full assessment. *Specificity*, also known as the *true negative rate*, is the proportion of cases correctly identified at screening to be not stuttering 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 is when a stuttering child is erroneously identified at screening to not have the disorder; in other words, when a child really is stuttering but is not identified as such. The problem there is that when it finally becomes apparent that an error occurred and the child really is stuttering, it may be that the best time in life for stuttering treatment has passed

False positive identification is when a child is erroneously identified at screening to be stuttering. In other words, when a child really is not 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 with identifying early stuttering when they bring children to clinics, the matter can be restated in the following way if parents are thought of as a screening procedure. There is good reason to believe that parent identification of early stuttering is sensitive, with a high true positive rate. However, the specificity of parent identification of early 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>186</sup> Surprisingly, such an important topic has attracted almost no research, with apparently only one preliminary report from more than 20 years ago.<sup>187</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 normal 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>188</sup> in the form of what is known as the Continuity Hypothesis. In effect, this idea is that stuttering and normal disfluency are not categorical things, but lie on either ends of a continuum. In other words, stuttering is an extreme form of normal disfluency:<sup>188</sup>

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)

That proposition proved to be rather controversial, with an experiment shortly after purporting to show that it was wrong,<sup>189</sup> and that stuttering normal disfluency in fact were “two reliable and unambiguous response classes” (p. 691). There was disagreement about the experiment,<sup>190,191</sup> and some years later another experiment came to the opposite conclusion.<sup>192</sup>

The Continuity Hypothesis still sometimes appears today in peer-reviewed clinical journal publications. For example, “parents learn which types of disfluencies are typically associated with childhood stuttering ... and which disfluency types are typically associated with normally (dis)fluent speech (p. 121).<sup>193</sup>

*The Diagnosogenic Theory*

Another historical influence promoted the idea that stuttering and normal disfluency lie on a continuum. This was Wendell Johnson’s extremely influential Diagnosogenic Theory—now defunct—that implicated normal disfluency in the cause of stuttering. The theory is considered in detail during Lecture Three, but, in short, it stated that stuttering emerged from parents believing that their children’s normal disfluency was stuttering.

### **Disorders to distinguish from stuttering**

There are some disorders that are broadly similar to stuttering, although not in the sense that they resemble each other and require a challenging differential diagnosis process to identify. Identifying them is straightforward with a case history and basic clinical observation. Their case histories and clinical features are obviously different from the disorder described so far during this lecture, which is sometimes referred to with the term *developmental stuttering* to distinguish it from the disorders that are now overviewed. The first task for a clinician when meeting new clients is to confirm the diagnosis of developmental stuttering rather than one of the following disorders, 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>194</sup> A recent guide to terminology for acquired stuttering<sup>194</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, and *psychogenic stuttering* refers to “a dysfluency that is somehow associated with a psychological problem or an emotional trauma” (p. 42).<sup>194</sup> Reports from the latter category, although common, are the most clinically puzzling, and it possible that some or all of them are a combination of neurogenic and

psychogenic.<sup>195,196,197,198,199,200,201,202,203,204,205,206,207</sup> The most recent report about this matter<sup>208</sup> suggests a differential diagnostic procedure for neurogenic and psychogenic stuttering, and 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>209,210,211</sup> Its defining features are different from developmental stuttering and are apparent to clinical observation.<sup>209</sup> Neurogenic stuttering occurs anywhere in words, but stuttering occurs mostly at the start of words, as described previously. Neurogenic stuttering occurs regardless of the grammatical class of words, which also is not the case for stuttering. Those affected by neurogenic stuttering appear rarely to be anxious about it, which is the exact opposite of stuttering, as considered during Lecture Ten. Nonverbal superfluous behaviours are common with stuttering but rare with neurogenic stuttering.

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>212</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>213</sup> stated “difficulty initiating speech, often repeating ‘dadadada’ before finding her words” (p. 137).

#### *Cluttering*

It is possible that someone who has the rare speech disorder cluttering<sup>214,215,216</sup> could be mistaken for having stuttering. The features of cluttering<sup>214</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>217</sup> So, as well as someone with cluttering being mistakenly identified as stuttering, it is possible for someone to have both disorders and for stuttering to be overlooked.

#### *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>218</sup> Such confusion is most likely to occur when tics have a vocal component. Motor—that is nonverbal—tics will occur when people are not speaking, and that does not happen with the superfluous behaviours of stuttering. The most common 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. It commonly is associated with psychological problems. A recent report suggested that as many as one fifth of children with Tourette Syndrome may have speech that resembles stuttering.<sup>219</sup>

#### *Neurological disorder*

When extremely severe cases of stuttering develop suddenly during early childhood the disorder may be mistaken for a neurological disease. Doctors have been known to refer cases of severe early 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>220</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. 1). 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' disease, and who were reported to be stuttering afterward.<sup>221</sup>

### An unusual case history

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, and potentially is comorbid with one or more of the disorders just described.<sup>†</sup>

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 syllable repetition and incomplete syllable repetition and nonverbal superfluous behaviours of muscle contractions round the 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.

## GUIDELINES FOR INTERACTING WITH THOSE WHO STUTTER

### An important topic

There is no shortage of recommendations to the public about how to interact with those who stutter.<sup>222</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 who 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>222</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)

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"
- (2) "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 ten respondents, in this rank order:

- (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:

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

- (1) "finishing ones 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 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." This is a justifiable concern, considering that extended stuttering moments may elicit unusual eye contact patterns. 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 the disorder 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>223</sup> Sometimes those 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, the explanation for that is a change to customary speech while on stage: louder, slower, perhaps with a different accent, and so on.

#### *The Modified Vocalisation Hypothesis*

There are some well-known changes to the customary way of speaking that are not so subtle. The term "fluency inducing conditions" is attributed to Wingate who proposed a Modified Vocalisation Hypothesis to explain why they reduce stuttering.<sup>224</sup> Wingate proposed that all fluency inducing conditions can be explained because "speaking under all of these conditions emphasizes vocalization and continuity of vocalization" (p. 682).<sup>224</sup> There is much research about these conditions, which is outlined in a reference text.<sup>225</sup>

#### *Singing*

Arguably the most commonly known feature of stuttering is that it goes away during singing. There are some who question whether this always happens (p. 425),<sup>226</sup> but it is generally accepted as a feature of the disorder. 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>227</sup> showed changes consistent with a popular treatment method to be discussed shortly: speech restructuring. In short, singing stabilises and simplifies speech motor activity.

#### *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. Demosthenes, the famous orator, lived during the third century BC. According to Plutarch writing in 75 BC, Demosthenes stuttered and consulted the Greek actor Satyrus who, among other treatments, prescribed that Demosthenes run or walk uphill while speaking.<sup>228</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 just simply dubious. An example of the latter is the now infamous "stuttering schools" that proliferated in the United States during the first part of the 20<sup>th</sup> Century.<sup>229</sup> Miniature in-the-ear metronome devices emerged during the 1970s<sup>230</sup> but never attained any demonstrable success.

#### *Reduced speech rate*

Virtually everyone who stutters will report at some time being told to "slow down." That advice is presumably based on an assumption by casual observers that the problem with stuttering is 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>231</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%, and left 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 stranger, if someone who stutters says what someone else has just said during a spontaneous monologue, but a few words later, the same thing happens. 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>232</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 was a series of laboratory experiments showing, in short, that if those who stutter receive electric shock or a loud noise after stuttering moments, then their stuttering decreases, and in some cases stops altogether, only to return when the shock or noise stops. There are at least 50 publications to that effect, dating from the early 1960s.<sup>233,234,235,236,237,238,239</sup> The shock or noise is called response contingent stimulation of stuttering. The results of those experiments show that stuttering has operant features.

#### *A disorder with 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 freely emitted problem behaviour. As discussed during Lecture Three, it is a physiological problem 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>240,241,242</sup> Research showing that this was an option for children<sup>243,244,245</sup> established clinical possibilities that have been fruitful, particularly for treatment of early stuttering, as discussed during Lectures Six and Seven.

There is some evidence, albeit patchy,<sup>246,247</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>248,249</sup>

### **Auditory feedback**

#### *Delayed and altered auditory feedback*

These terms 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 similar to a hearing aid. The first such effect to be discovered was delayed auditory feedback,<sup>250,251</sup> often referred to as DAF. Subsequent to a famous report of it being used to reduce stuttering,<sup>252</sup> this discovery profoundly influenced treatment practices. Generally, delayed auditory feedback creates a slow and unusual drawling speech pattern that reduces or eliminates stuttering. 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. Those treatments are referred to generically as speech restructuring.<sup>253</sup>

For nonstuttering speakers, delayed auditory feedback can induce disfluencies that once were thought to resemble stuttering,<sup>254</sup> and this prompted many theories 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>226</sup> inducing transient speech problems (p. 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 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.

## **SUMMARY**

The disorder of stuttering can be associated with potentially confusing terminology that is best to avoid. It is a clinically useful idea that the disorder involves moments of stuttering 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. Identifying stuttering is generally not a clinical challenge, with adults and parents usually being correct with their identifications. However, there are some disorders that potentially could be mistaken for stuttering. 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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#### HOW STUTTERING AFFECTS PEOPLE

##### 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 during Lecture One 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>1</sup> when given a spontaneous speaking task, on average those who stutter say around one third less than those who do not stutter. A more recent publication<sup>2</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 than a quarter of peers. So a person severely affected for a lifetime may say only a quarter of what is possible, or take four times as long as others to say what is intended.

###### *Word avoidance*

It is well known that those who stutter may attempt to limit its impact on daily life by avoiding words.<sup>3,4,5</sup> Scanning ahead for words that are difficult, and avoiding them with circumlocutions, is a common strategy. However, there are some words that cannot be avoided. Examples are your name, telephone number and address, and a destination to which you wish to travel on public transport.

###### *Grammatical constraints*

There have been reports that those who stutter may attempt to make the best of their speech output by restricting their use of grammar.<sup>2,6</sup> The latter of those 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>2</sup> A particularly noticeable reduction of modality was 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>7</sup> The authors concluded that those who stutter have “a reduced openness to interpersonal engagement within communication exchanges” (p. 1). A follow-up report of the participants in that study after speech treatment<sup>8</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>9</sup> showed these treatment gains to be maintained, with evidence of continued improvement. There is a suggestion from a study<sup>10</sup> of eight 7-year-old children who stuttered and eight control children that such problems may begin early during the course of the disorder.

##### 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>11,12,13,14,15</sup>

###### *Measurable impact*

The quality of life impairment that stuttering can cause is well demonstrated by a film depicting the life of King George VI.<sup>16</sup> United States Vice President Joe Biden suffered from early stuttering and described the disorder as “the single most defining thing in my life.”<sup>17</sup> This is despite his wife and

daughter being killed in a car accident, and being elected Vice President. Two studies<sup>15,18</sup> used a medically oriented quality of life instrument to show that stuttering participants had poorer quality of life compared to controls. The former study also reported that stuttering affects quality of life as adversely as life threatening conditions such as neurotrauma and coronary heart disease. Presumably that is because, in contrast to those diseases for the most part, it is present across the lifespan.

A study<sup>19</sup> recruited 78 participants, four of whom stuttered, to a “willingness to pay” and “quality adjusted life years” analysis of the disorder. The nonstuttering participants were provided with detailed information about stuttering. Results indicated that participants would pay “with amounts of money equal to two to four times their annual incomes” (p. 309)<sup>19</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>19</sup> Those results were consistent with quality of life impairments measured for serious medical illnesses.

Bandura’s<sup>20</sup> notion of self-efficacy refers to the extent to which people believe they will be able to achieve things. For those who stutter, a report<sup>21</sup> of 39 adults found a relationship between high levels of self-efficacy and positive quality of life. The effect was found to occur independent of stuttering severity.

#### *Stuttering across the lifespan*

After the pre-school years stuttering seems to worsen throughout life. This is apparent from comparing data sets that measure stuttering severity during the pre-school years and during adulthood.<sup>22</sup> There have been suggestions that stuttering decreases in prevalence and severity with older age.<sup>23,24</sup> However, the majority of studies suggest that stuttering, and the various problems associated with it, do not abate with advancing age.<sup>25,26,27,28</sup>

### **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>29</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>30</sup>

#### *Stuttering impairs occupational attainment*

It is not surprising, then, that stuttering has an impact on occupational attainment. Those who stutter say that this is the case, with one report showing that 70% of 200 stuttering adults thought the disorder prevented promotion, and 20% declined the challenge of a promotion because of it.<sup>31</sup> One report<sup>32</sup> even indicated that 7.5% of participants had employment terminated because of their stuttering. Another report indicated that speech rehabilitation resulted in improved occupational level and promotion prospects.<sup>33</sup> The disorder appears to affect everyday experiences in the workplace.<sup>34,35</sup> Employers have reported that those who stutter are less employable and promotable than others.<sup>36</sup> Members of the public seem to reflect those attitudes.<sup>37,38</sup> Study of a birth cohort<sup>39</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.

### **Educational impact**

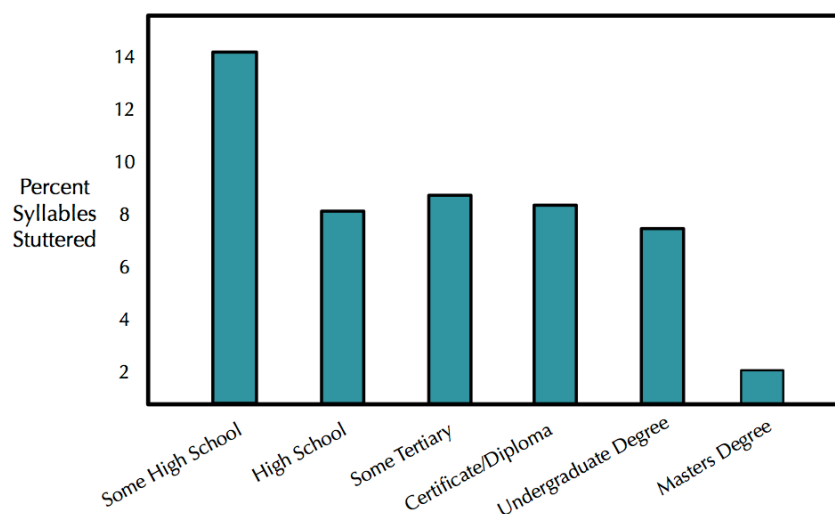
#### *The school years*

Some early publications identified education problems for children who stutter during the school years,<sup>40,41</sup> and those results have been replicated in a more recent report.<sup>42</sup> A compelling, large cohort,<sup>43</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. Those educational problems are not explained by lower intelligence of those who stutter, with a review of 13 studies<sup>125</sup> showing no evidence of that being the case.

### *High school onwards*

The birth cohort study mentioned earlier<sup>39</sup> reported that stuttering had no effect on educational outcome. However, there is evidence that the disorder has a negative impact on education attainment. One report was a negative linear relationship between stuttering severity and education attainment.<sup>44</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 diagram.<sup>45</sup> The vertical axis has 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 masters degree at university.



One report<sup>46</sup> produced a troubling statistic 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 point 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.

## **Stuttering stereotypes**

### *Generally*

Many research publications have shown that those who stutter are affected by negative stereotypes about their disorder.<sup>47,48,49,50,51,52</sup> This appears to be true across the lifespan from childhood to adulthood.<sup>53,54,55</sup> The topic was considered to be of sufficient importance to warrant a conference about it.<sup>56</sup> Stuttering is frequently depicted in movies and television, and more often than not with a negative stereotype.<sup>57</sup> A review of 29 works of fiction that contained a character who stuttered<sup>58</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>59</sup> shows it typically to be used in a negative fashion, such as a comic device or as a sign of weakness.

A report<sup>60</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 from the United States,<sup>61</sup> two thirds of whom were men, involved a list of 15 stigmatising experiences. For most of those experiences, the majority of participants reported them having occurred during their lifetimes. Interestingly, “most participants reported experiencing them never or rarely in the past year” (p. 16).

There is compelling evidence that communities of various cultures hold negative stereotypes about those who stutter. A review of that evidence<sup>62</sup> states

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 that review, publications have reported stuttering stereotypes in United States,<sup>63</sup> Polish,<sup>64</sup> and Chinese populations.<sup>65</sup> As noted earlier, that stereotyping extends to occupational suitability.<sup>37,38,63</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>66</sup>

Added to that literature is a recurring finding over several decades implicating speech-language pathologists in the perpetuation of negative stereotypes about the disorder. Clinicians in the United States,<sup>67,68</sup> the United Kingdom,<sup>69,70,71</sup> Korea<sup>72</sup> and Turkey<sup>73</sup> are reported to have negative and empirically unjustifiable attitudes to stuttering, although that research shows some signs of improvement of that problem over time.

A long-standing project has used a tool called the *Public Opinion Survey of Human Attributes-Stuttering* (generally referred to as POSHA-S) to explore worldwide attitudes to the disorder. A summary of a series of European reports<sup>74</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. Subsequently, reports dealing with attitudes in Turkey<sup>75</sup> and Portugal<sup>76</sup> have been published. A report with that database<sup>77</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>78</sup> reported that awareness and accurate knowledge about stuttering was limited in Japan.

A report<sup>79</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 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 about a disorder. 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.

### Adolescence

During adolescence, it seems that peers find those who stutter “nervous” and less attractive than others.<sup>80,81,82,83,84</sup> One of those findings<sup>84</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.

### Teachers

Some reports suggest that United States teachers of children younger than 12 years,<sup>85,86,87</sup> special educators in schools,<sup>88</sup> and school administrators<sup>89</sup> may well hold negative and unjustifiable stereotypes about the disorder. More recent reports from the United States<sup>90</sup> and India<sup>91</sup> were more favourable, although another report showed that United States teachers did not have more accurate beliefs about stuttering than the general public.<sup>92</sup>

A recent report<sup>93</sup> of 262 Kuwait teachers and 209 trainee teachers was rather sobering. The report found, among other things, that 81% of the teachers and trainee teachers believed stuttering to be caused by emotional problems, 76% believed it to be precipitated by “a very frightening event,” and 15% believed that “a virus or disease” (p. 60)<sup>93</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>93</sup> A follow-up study<sup>94</sup> showed that (fortunately) such stereotypes could be corrected during teacher training with a 17-minute educational video about stuttering. A report of interviews with 10 Belgium teachers of adolescent students<sup>95</sup> showed that they felt stuttering could become a problem if attention is given to it, and that they tried to minimise reaction to stuttering and rarely talked about it in class.

There is some evidence that stereotypes about stuttering extend to university environments. University students have been shown to have negative attitudes to the disorder.<sup>96,97</sup> One report<sup>98</sup> showed that university professors and students scored students who stutter as having more negative personality traits than other students. However, another report<sup>99</sup> of student perception failed to find overriding stereotypes, and another found “neutral to positive perception” (p. 206)<sup>100</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>101</sup> A report of students from an Australian university<sup>102</sup> indicated a positive attitude toward stuttering, and suggested a connection between that result and curriculum content.

### Guidelines Anticipation of stuttering

#### *A common effect*

It has been known since the 1930s that those who stutter anticipate its occurrence with some reliability.<sup>103,104,105,106,107,108</sup> That knowledge has been bolstered by reports with adults<sup>109,110,111,112,113</sup> and children<sup>114,115,116</sup> during reading tasks that have established eye gaze patterns consistent with anticipation of difficulty with certain words. That knowledge prompted many of the influential causal theories in the history of thought about the disorder (see Lecture Three): primary and secondary stuttering theory, the Diagnosogenic Theory, approach-avoidance theory, and the Anticipatory Struggle Hypothesis.

#### *The experience of anticipating stuttering*

A recent study of 30 adults<sup>5</sup> reported their experiences of anticipating stuttering, and around half reported “they experience anxiety or uncertainty when they anticipate stuttering” (p. 18). 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. 13) was reported by 87% of them. Circumlocution and substituting an alternative response during conversation—not the one 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>117</sup> Avoiding situations was also a common proactive response to anticipating stuttering.

“Self management strategies” (p. 15), either learned in a clinic or self generated, were reported by 87% of participants. Those included variants of the speech restructuring technique overviewed during Lecture One, 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 and 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% reporting that it is always helpful.

Research has begun<sup>118</sup> to develop an instrument to measure anticipation of stuttering events: the Premonitory Awareness in Stuttering Scale. This 12-item scale was adapted from a similar scale used for tics, and showed that adults who stuttered reported anticipation of “speech disruptions” more often than control speakers.

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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.

## Social anxiety

### *Situation avoidance*

The effects of stuttering on people in the ways just discussed—their 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 discussed then, a common effect of social anxiety is to avoid speaking situations.

### *Situations commonly avoided*

An early report<sup>119</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 table<sup>‡</sup> presents the top 15 situations that were avoided by the groups, with the most avoided ones 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 seems also that those who stuttered avoided group speaking situations more often than controls. The report showed also that those who stuttered were the most comfortable with people they knew, such as friends and family, and did not commonly avoid those situations.

Things seem not to have changed during the decades since the first report. A more recent publication<sup>120</sup> used another situation checklist<sup>121</sup> with 88 adult participants seeking treatment for

<sup>‡</sup> 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.

stuttering and 209 controls. The report documented the troublesome speaking situations that best distinguished between the groups. The top 10 situations are presented in the table, in order of their “strength of the association ... to negative emotional reaction” and the top six situations are presented in order of their “being associated with the likelihood of speech breakdown.”

NEGATIVE EMOTION	SPEECH BREAKDOWN
“Talking on the telephone”	“Talking on the telephone” or Returning a phone call”
“Asking a teacher or supervisor a question”	“Talking with teachers or supervisors”
“Trying to get your point of view across”	“Saying a sound that previously has been troublesome”
“Being rushed”	“Chatting with friends”
“Chatting with friends”	“Giving your name”
“Trying to make a good impression”	“Making an appointment”
“Saying a sound or word that previously has been troublesome”	(p. 19–20) <sup>121</sup>
“Being asked to give personal information”	
“Telling a taxicab driver where you want to go”	
“Introducing yourself”	
(p. 19) <sup>121</sup>	

### *The telephone*

The problematic nature of 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. One report<sup>122</sup> was a survey of 223 British participants. 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 that 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” (p. 308–309).<sup>122</sup> Recurring reasons given for this were that nonverbal communication is not possible by telephone, reactions to stuttering are unknown on the telephone, 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>123</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>122</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>124</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>125</sup> reviewed 33 studies dealing with the personality of those who stutter during the period 1928–1985 (Table 7-1, p. 193), and 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. 209)

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>126</sup> reported significant differences between the two groups. Another report<sup>127</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 normal range for the five domains, but had higher Neuroticism and lower Agreeableness and Conscientiousness scores than controls.

Another report<sup>128</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>129</sup> was consistent with the Neuroticism finding by reporting that it correlated with poor quality of life among those who stutter.<sup>†</sup> However, using the same measure with a culturally different population, another study<sup>130</sup> found only higher Agreeableness scores for the stuttering group, but no other differences.

Considering the inconsistent and often slight differences found across studies, it seems obvious that the personality differences of those who stutter as a group are a reflection of the effects of having the disorder for a lifetime rather than the nature or the cause of the disorder.

## 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>131</sup> and compact reviews<sup>132</sup> are available, along with an overview of the current status of the field.<sup>133</sup> More technical overviews are available<sup>134,135,136</sup> with more focus on the science of genetics, and there is an overview of specific genetic mutations identified to date.<sup>137</sup>

Clients and their parents who don't know it already can be relieved to hear that genetics is involved with the condition, rather than it being a psychological problem, as suggested by common stereotypes discussed earlier. Clinically, it can also be useful to introduce a discussion of what causes stuttering by stating that genetics are involved with it.

The first review paper<sup>131</sup> outlines the progress with accumulating knowledge about the genetics and stuttering in four methodological phases: familial incidence, twin studies, family aggregation, and biological genetics. The following overview follows those headings.

### Familial incidence

*Family history is common*

Fundamental evidence for genetic involvement with a disorder is vertical transmission: in other words a family history. The review mentioned earlier<sup>125</sup> of 21 reports dating from 1937 to 2005 (Table 3-5, p.

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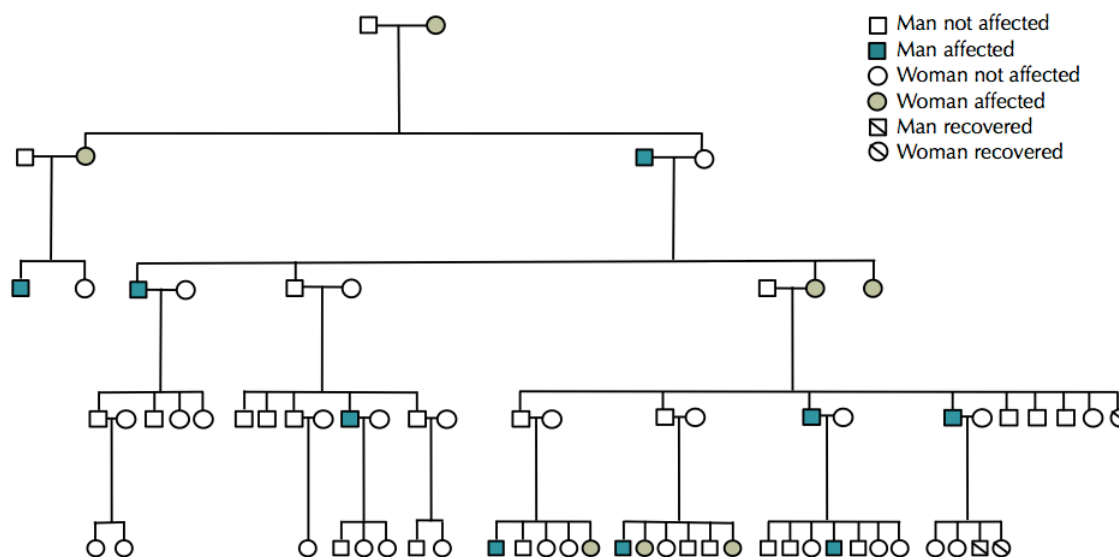
<sup>†</sup> Neuroticism is a tendency to experience high levels of unpleasant emotion such as anxiety, anger and sadness.

95) shows that, overall, 69% 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, however, to believe that such participant-report data underestimate the true family history rate, with many family members with affected relatives failing to report it.<sup>206</sup> So clinically, if an interview suggests that there is no family history, there is some room for doubt.

#### *A famous family*



The diagram<sup>‡</sup> is from a 1940 report of five generations of an Iowa family in the United States.<sup>138</sup> Clinicians routinely see many families during clinical practice with stuttering running 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.

## Twin studies

### *Identical and non-identical twins*

Twin studies are another way of establishing a genetic basis to a disorder. 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 disorder it is referred to as concordance, and if only one twin has a disorder it is referred to as discordance. So, if monozygotic concordance for stuttering is higher than dizygotic concordance, that strongly suggests that genetics is involved with the disorder. In other words, if identical twins both stutter more often than non-identical twins, that suggests genetic involvement. (The assumption underlying that reasoning is that the living environments of both types of twins are the same.)

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

It seems to have been discovered in the 1930s that monozygotic concordance for stuttering is greater than dizygotic concordance,<sup>139,140</sup> with the first attempt to quantify concordance rates some years

<sup>‡</sup> 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.

later.<sup>141</sup> In 1981 a seminal study of 30 twins<sup>142</sup> was published and its findings were subsequently replicated with larger cohorts.<sup>143,144,145,146,147,148</sup>

A large parent self-report study of 10,500 5-year olds<sup>148</sup> found, for “probable stuttering,” dizygotic concordance for boys 36% and 34% for girls, and monozygotic concordance 53% for boys and 61% for girls. That report estimated “probable stuttering” to be 42% inheritable; in other words a 42% genetic contribution to the disorder. The highest estimate of genetic contribution to stuttering was a self-report study<sup>147</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, twins who are genetically identical are more likely to both stutter than are non-identical twins. The broad interpretation of those studies is that 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 disorder is transmitted genetically.<sup>131</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>149,150</sup> were the first application of the family aggregation method with stuttering, followed after two and a half decades by the first comprehensive report.<sup>151</sup> The latter study involved 213 people who stuttered, 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.

#### *A take-home message*

After more than a decade, Kenneth Kidd and colleagues at Yale University added to those family aggregation data,<sup>152</sup> and subsequently published data about 600 probands until the mid 1980s.<sup>153,154</sup> A take-home message was provided by pooling data for families covered by the first comprehensive report<sup>151</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>155</sup>

One common interpretation of that information is that women pass on the genetic material for stuttering more than men do, but are themselves affected less often than men.

#### *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 developing the disorder by “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 verified that such concerns are not justified<sup>156</sup> by showing no association between birthrank 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 result would have been expected from the Yale data. That result is consistent with what is currently known of the nature and cause of the disorder, as discussed during Lecture Three. There is no reason for parents to be concerned that stuttering can be transmitted by “copying.”

## Biological genetic evidence

Direct genetic evidence is another technique to explore the genetics of stuttering. Until recently, the most up-to-date procedure was genome wide linkage studies, which trace patterns of stuttering inheritance through generations using genetic markers. The review paper mentioned earlier<sup>132</sup> lists six studies that have provided evidence of multiple chromosome linkage.<sup>157,158,159,160,161,162</sup> Those studies make it seem probable that the disorder 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>131</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 disorder. To date, this method has been reported in one study about stuttering, which again suggested polygenic inheritance.<sup>163</sup>

Another source of biological genetic evidence with the disorder is candidate gene analysis, which provides information about contributions of specific genes. Reports using this method have identified mutations in several genes to date (GNPTAB, GNPTG, NAGPA, AP4E1).<sup>164,165,166,167,168,169,170</sup>

## A mouse model of stuttering

Considerable interest has been generated recently with an (arguably adventurous) attempt to establish a mouse model of stuttering.<sup>171</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 reported also for the experimental mice. The researchers concluded that their results established “the mouse as an attractive model for studying this disorder” (p. 1009).<sup>171</sup>

## Conclusions

### *A recent cell biology focus<sup>†</sup>*

Recent directions of genetic research about stuttering have focused exclusively on cell biology. All gene mutations identified to date 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 involved are associated with stuttering, and severe mutations are associated with serious disorders including neurological disease involving intellectual disability and white matter pathology.

Beyond that information, recent genetic research with stuttering has not dealt with the disorder in a broader context. Recent publications have not proposed any links between established gene mutations and the many well know features of stuttering—outlined during the next lecture—that need to be accounted for when trying to understand its cause. In particular, those reports have not attempted to deal with the anomalies of brain structure and function associated with stuttering that will be discussed shortly. Also, there has been no proposed link between the gene mutations identified and the fact that stuttering affects more boys than girls, and the fact that the same occurs with other brain based disorders, such as language disorder and autism.

### *Almost certainly a polygenic disorder*

Part of understanding stuttering is to know what is necessary and sufficient<sup>†</sup> for it to occur.<sup>172</sup> It is obvious that genetics is neither of those things. Around 70% of those who stutter report a family

<sup>†</sup> This content was adapted from material supplied by Marcus Hinchcliffe, Department of Medical Genomics, Royal Prince Alfred Hospital, Sydney.

<sup>†</sup> Chapter One of that text, dealing with necessary and sufficient conditions for establishing cause, is recommended reading in the context of understanding stuttering. The concept of necessary and sufficient conditions for stuttering causality occurs many times during these lectures.

history, so genetic involvement is not always present, meaning that genetics is not necessary for stuttering. And twin studies make it obvious that genetics does not always lead to stuttering, so it is not sufficient for stuttering. Even though twin studies support estimates that stuttering is around 80% inheritable, genetic mutations have been discovered that can explain only 10% of stuttering occurrences, with many of those mutations uncovered in stuttering-dense consanguineous families.

It is clear that the genetics of stuttering is complex, with incomplete penetrance within families, and some rare individuals with mutations of a single gene as a major contributor to the disorder. All this makes it virtually certain that stuttering is a polygenic disorder, caused by combined actions of more than one gene, such as with hypertension and coronary heart disease.

## BRAIN STRUCTURE AND FUNCTION

### Mounting evidence

There is a large and growing body of evidence during the past decades showing that children and adults who stutter have anomalies of brain structure and function in areas subserving spoken language. That research literature expands each year. A 2017 edition of the *Journal of Fluency Disorder* was assigned specifically to this topic. A review of neuroimaging studies<sup>173</sup> found more than 100 reports during the period 1995–2016. That research incorporates evidence of unusual structural and functional non-dominant—right sided—brain activity in speech areas. Additionally, there is evidence that change of brain function occurs after brief behavioural stuttering treatments.<sup>174,175,176,177,178</sup> The more recent studies suggest that the problem is one of connectivity; transmission of information along the white matter fibres of the brain is impaired in areas involving spoken language. White matter fibres form complex connections between executive areas of the cortex, and are critical for the development of complex neural networks needed for spoken language. Neuroimaging studies of stuttering continue to accumulate rapidly, and overviews and meta-analyses of that body of research are available.<sup>173,179,180,181,182</sup> An overview of the current status of the field is available.<sup>133</sup> There is a report<sup>183</sup> that grey matter of the brain is affected also in adults, with a stuttering group having a larger right nucleus accumbens—a subcortical structure—than a control group.

An essential context for this information, however, is that it is not constrained to the disorder of stuttering. For example, anomalies of brain structure and function, or both, have been reported to be associated with specific language impairment<sup>184</sup> and speech sound disorder.<sup>185</sup>

### Two current hypotheses

The arcuate fasciculus is a 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. There are now two reports of adult and control stuttering participants linking structural abnormalities in the arcuate fasciculus with stuttering.<sup>186,187</sup> The researchers in the latter study reasoned that "because these structures emerge during the second trimester of embryogenesis ... it seems reasonable to speculate that the unique changes in arcuate fasciculus ... might also arise during this period (p. 273).<sup>187</sup>

Some researchers<sup>188</sup> have drawn on the results of neuroimaging research and genetic research implicating lysosomal metabolism, as previously discussed, to present a testable hypothesis: The onset and development of early childhood stuttering is linked to abnormal or late myelogenesis of perisylvian fibre tracts.

### The critical issue

It is obvious that brain anomalies, alone, are not sufficient for stuttering to occur. Arguably the fact that monozygotic concordance for stuttering is not 100%, as discussed earlier, is the clearest

demonstration of that. It is also 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 discussed shortly, stuttering does not occur when children first start to speak, but occurs some time after during early language development. If brain anomalies are somehow fundamental to the disorder, something additional must occur for stuttering to appear.

What is yet to be determined, though, is whether brain anomalies are necessary for stuttering to occur: whether those anomalies are always present when stuttering is present. And even if brain anomalies are necessary for stuttering, it needs to be determined whether they are part of the cause of stuttering or are a consequence of it.<sup>189,190</sup> That issue does not pertain specifically to stuttering. It has, for example, been discussed in relation to developmental language disorders.<sup>191</sup> Some support—but not conclusive evidence—for 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>192,193,194,195</sup> and similar results for younger children 3–10 years old.<sup>196,197,198</sup> Neuroimaging studies of children have been overviewed.<sup>199</sup> The latter report,<sup>198</sup> published after that review, argued that the varying location of brain differences between stuttering children and controls might be caused in part by the inclusion of children in the former groups who eventually recovered naturally. Consequently, the authors conducted a study to take account of that issue with 3–10 year-olds. They reported that, compared to control children, the children with persistent stuttering had slower growth rates in the arcuate fasciculus and corpus callosum (the corpus callosum is a large white matter fibre structure connecting the two hemispheres of the brain). However, that was not the case for the children who recovered from stuttering.

Ultimately, the only way to resolve the issue of whether brain anomalies are necessary for stuttering and are involved causally is to do brain scans of infants who are genetically at risk of stuttering. Such studies are currently under way and, if successful, will determine whether children who develop stuttering have brain anomalies that are present before the developmental appearance of the disorder. That will exclude the possibility that the brain anomalies identified so far have resulted from several years of stuttering. In the report mentioned previously,<sup>198</sup> for example, the children in the persistent stuttering group had a mean age of 7 years 10 months.

### **Clinical applications of neuroimaging research**

Ideally, the benefits of research about brain structure and function with stuttering will eventually improve treatment for the disorder.<sup>200</sup> In the interim, those authors 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. 11). Indeed, that research is destined eventually 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.

## **STUTTERING EPIDEMIOLOGY**

### **Epidemiology**

Epidemiology 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 designs do not attempt any intervention, and common methods are cross-sectional studies, cohort studies, or case-control studies. Common experimental studies that test interventions in populations are randomised controlled trials or quasi-experimental designs.

In the case of stuttering, the favoured methods are observational. There has been no epidemiological study of a stuttering intervention, and 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>201,202</sup> with eventual consequences of a public apology from the university concerned and legal compensation to the participants decades later.

### 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 the disorder is and 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 disorder occurs frequently, causes significant distress, and is successfully treatable—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>203,204</sup>

## POINT PREVALENCE OF STUTTERING

### Point prevalence

Point prevalence of a health issue or disorder, 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 disorder is 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>132</sup> details all the caveats that need to be kept in mind by the consumer of research 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 affected or not. But for practical reasons more than anything else, mostly that does not happen, and most of the available stuttering point prevalence information comes from reports given by relatives, or by self-report.

Although, as discussed during the previous lecture, 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>205</sup> and another found the opposite.<sup>206</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 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.

Telephoning households to find people who stutter<sup>207</sup> seems at first thought to be a way around this problem. However, as also discussed later during this lecture, there is good reason to believe that those who stutter may avoid speaking on the telephone.

### Estimates of stuttering point prevalence

A well-known reference text<sup>125</sup> documents 46 international cross-sectional studies from 1893 to 2006 (Tables 3-1 and 2-1, p. 79–80) dealing with school children. The mean reported point prevalence in those tables is 1.2%. However, the standard deviation is quite large at 0.9, because the 46 estimated values vary considerably. The lowest reported prevalence figure is 0.3 and the highest is 5.2, suggesting that either the point prevalence of stuttering varies from country to country, or that the variation represents errors with estimating prevalence across the studies.

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 cultures.<sup>208</sup> Stuttering occurs in both languages with bilingualism.<sup>209</sup> That being said, the review article mentioned previously<sup>132</sup> notes that one recent study with many participants (N=119,367)<sup>210</sup> convincingly 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>211</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>212</sup>

That review article<sup>132</sup> presents 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 here.<sup>‡</sup>

PARTICIPANTS	AGE IN YEARS	PREVALENCE	MALE/FEMALE RATIO	
1,113 <sup>213</sup>	4–5 [1]	2.2	0.7:1	
4,983 <sup>214</sup>	4.5	5.6	[3]	
3,165 <sup>215</sup>	2–5	2.6	2.6:1	[1] Not clear in the report but this probably is the age range
10,000 <sup>216</sup>	5–13	0.3	[3]	
21,027 <sup>217</sup> [2]	6–10	0.8	5.1:1	[2] The study reports data separately for the different age groups
	11–15	0.5	4.7:1	
	16–20	0.3	1:9:1	[3] Not reported
12,131 <sup>207</sup>	2–99	0.7	2.3	[4] For the entire sample
119,367 <sup>210</sup> [2]	3–10	2.0	2.5 [4]	
	11–17	1.2		

<sup>‡</sup> 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.



As the authors of the review article note, their conclusion is consistent with the occurrence of natural recovery after onset, as discussed shortly. The conclusion is also consistent with early stuttering being particularly responsive to treatment compared to later periods of life, as discussed during 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 offered that suggestion.<sup>218</sup>

### **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>210</sup> which is the principle 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 had “a doctor or health professional” (p. 1035)<sup>210</sup> ever told them 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 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 report, stuttering was the equal ranked third most prevalent.

But still, the results from that study might be an underestimate, regardless of any merit with its methods. One reason is that a minority of parents of pre-school children with communication disorders seem to seek health care advice about such disorders.<sup>219</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 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.

## **CUMULATIVE INCIDENCE OF STUTTERING**

### **Cumulative incidence**

Cumulative incidence, sometimes referred to simply as incidence, is the number of new cases of a disorder during a certain period. It does not include recoveries during that period. So for example, the cumulative incidence of a disorder up to 12 years of age remains the same regardless of how many recover from the disorder. 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 disorder is with prospective epidemiological methods, which study its developmental course with a cohort of children. In the case of a disorder such as stuttering, however, a study of natural development is not without methodological problems. 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 stuttering.<sup>†</sup> There have been three longitudinal studies published for stuttering, which are now discussed.

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

*The 1,000-family study*

A prospective study of a cohort of children published in 1964,<sup>220</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 table incorporates information provided at another source<sup>221</sup> (p. 10) about the numbers of children in the cohort at various ages, and the data in Figure 3 (p. 32) of the original publication<sup>220</sup> about the numbers of children who stuttered. As mostly occurs with longitudinal studies, the participant numbers decrease over time, which is known as participant attrition.

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

Albeit a landmark study and decades ahead of its time, the 1,000-family study methods of stuttering identification are currently recognised as a serious weakness that damages the credibility of its results by modern standards.<sup>132,222</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>223</sup> involved all 1,042 children born during 1990 and 1991 on the Danish island of Bornholm. The health services of that island included “a free speech and hearing evaluation” (p. 49)<sup>223</sup> by a clinician. 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 people, such as nurses, social workers, and teachers, who were in position to know about the children” (p. 51).<sup>223</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>223</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, for an 8-year cumulative incidence of 5.2%. The 12-year cumulative incidence remained unchanged at 5.2%. Of the children who stuttered, 52% were boys and 48% were girls.

The authors of the review article mentioned previously<sup>132</sup> describe a subsequent Danish publication from Bornholm<sup>224</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.<sup>132</sup> (p. 71)

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

The children in this report are part of a cohort study of child language development in Melbourne, Australia: the Early Language in Victoria Study (ELVS).<sup>225,226,227</sup> The study is 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 this fridge magnet 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>228</sup> The parents were sent reminder letters about the study every 4 months for 12 months.

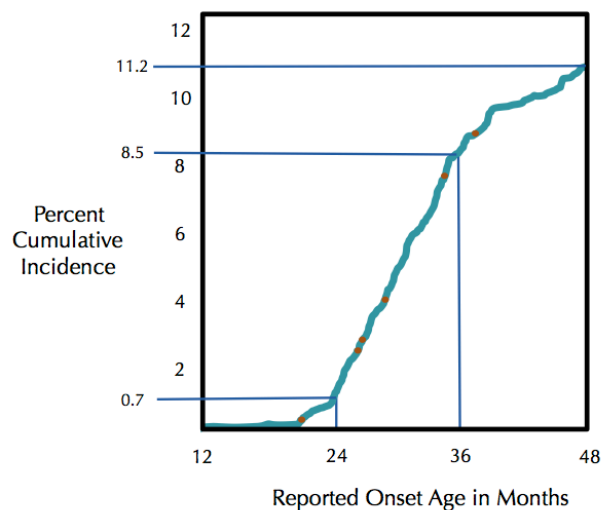
When a parent reported stuttering onset a clinician visited the home, recorded a case history, and made a video recording of the child's speech. By 3 years of age 158 parents reported stuttering onset. The visiting clinician confirmed the presence of stuttering for 137 children and was unsure about 21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

The first ELVS report<sup>228</sup> showed a 3-year cumulative incidence of 8.5%: 137 of 1,619 children. A subsequent report of the cohort 12 months later,<sup>229</sup> without any participant attrition, was a 4-year cumulative incidence of 11.2%: 181 of 1,619 children. That represents one in nine children. Of the children who stuttered at that time, 59% were boys and 41% were girls.

The graph<sup>‡</sup> 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 as the cohort is studied further into childhood.

#### *Conclusions about childhood cumulative incidence*

Perhaps the most cautious interpretation of the three prospective studies of the matter to date is that an exact figure for childhood cumulative incidence of stuttering has yet to emerge. The estimate of 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 an extremely prevalent disorder.



#### **Lifetime cumulative incidence**

The lifetime cumulative incidence of stuttering is the risk of being affected at some time during life, including transient periods. In the reference text mentioned earlier<sup>125</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 one time either consider themselves or are considered by their parents to be stutterers is at least as high as 10 percent. (p. 91)

The authors of the review article mentioned earlier<sup>132</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 the disorder at some time during life.

## **STUTTERING ONSET**

### **Onset occurs during the pre-school years**

There is a general consensus that stuttering onset occurs early during the pre-school years,<sup>125</sup> and that 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 reported onset had a reported mean onset at 30 months.<sup>230</sup> Another study reported information about 22 children who stuttered, up to their fourth birthday, who had been stuttering for up to 1 year.<sup>231</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.

<sup>‡</sup> Adapted and reproduced with permission: Pediatrics, Vol. 132, Pages 460–467, 2013, © American Academy of Pediatrics.

These results are consistent with the ELVS report of 3-year cumulative incidence,<sup>228</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>229</sup> was 31 months.<sup>†</sup> Those results were consistent with a report of 87 children,<sup>233</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>232</sup> was mean onset 35 months with a range of 19–68 months.

A caveat to keep in mind here is that those reports were not designed to detect cases of stuttering that might occur towards the end of the pre-school years and later during the early school years, 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 so far that the bulk of stuttering onset occurs during the early pre-school years.

### **Onset can be sudden and severe**

An unusual feature of stuttering, unlike other speech problems such as phonological or language disorders, is that it appears after a period of normal speech development. This can be distressing for parents, particularly when stuttering onset is sudden. Reports show that half of cases start within 1 week and a third start during a single day.<sup>231,233</sup> The ELVS community cohort replicated those findings,<sup>228</sup> reporting that 50% of cases appeared during 1–3 days and 37% during a single day. It occurs sometimes that parents report a child going to bed speaking normally and at breakfast being severely affected by stuttering. There are reports of stuttering suddenly beginning during the course of an unremarkable day. One of many available case studies describes such sudden onset.<sup>234</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>230,235,236</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 the disorder described them as routine.<sup>237</sup> More modern reports substantiate their prominence at onset.<sup>238,239</sup> One study<sup>231</sup> reported nearly all of 22 parents stating that their children repeated whole words and syllables at onset, and another<sup>228</sup> reported 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.

### **More boys and men are affected than girls and women**

Proportions of boys at reported onset range from 50%,<sup>231</sup> 61%,<sup>228</sup> and 68%.<sup>233</sup> The ratio of men to women affected by stuttering ranges from 3:1 to 5:1 in various reports,<sup>125</sup> which translates to 75–83% men. There are two features of those figures to note. First, it seems that there are more boys and men stuttering than girls and women. Second, considering all publications about the matter, the reported ratios for pre-school children seem to be more evenly balanced between genders than are ratios for adulthood. So, in short, it seems that fewer girls begin to stutter than boys, and that they are more prone to recovery than boys.

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

## Is stuttering onset predictable?

### *Statistically*

From a statistical viewpoint, the ELVS prospective cohort study provided a few positive results.<sup>228</sup> At 3 years of age, being a boy, being a twin, having advanced vocabulary, and a mother with advanced maternal education, were significantly associated with stuttering onset. At 4 years of age the results were the same,<sup>229</sup> with two minor exceptions. Advanced vocabulary did not predict stuttering onset. However, scores for the Communication and Symbolic Behaviour Scales 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 United States National Health Interview Surveys showed that, along with many developmental disabilities, stuttering was associated with low birth weight.<sup>240</sup> The ELVS cohort had few twins on which to base a claim of a connection between twinning and stuttering onset. However, low birth weight is common with twins, so it might be that the ELVS data are consistent with the United States data.<sup>241</sup> However, a British study<sup>242</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 with reasonable participant numbers. One was a study of 87 stuttering pre-school children, mean age 39 months.<sup>233</sup> The researchers studied them retrospectively an average of 5.8 months after reported stuttering onset. A positive finding was consistent with the ELVS cohort study, that gender was associated with stuttering onset.<sup>†</sup>

Another report<sup>243</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 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 intervention 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.

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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.

## NATURAL RECOVERY FROM EARLY STUTTERING

### What is the natural recovery rate?

The next issue is how many children who begin stuttering during the pre-school years will recover naturally without needing 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.

### Two essential caveats

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

A complicating factor here is that there are grounds to believe that many parents do clinically useful things for early 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>220,244,245,246</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 during the previous lecture. As such, they may well be clinically useful things for parents to do.

#### *Natural recovery and treatment bias*

During such extended studies of children who stutter, it is not ethical for researchers to prevent parents of stuttering pre-school children from seeking treatment so that they can study the natural course of the disorder. 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 not available at present.

### Prospective reports of natural recovery beyond the pre-school years

#### *The largest cohorts*

The earliest prospective report of natural recovery was the 1,000-family study,<sup>220</sup> which indicated that 34 of the 43 children who stuttered (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>247</sup> which followed 84 children for 4 years post onset and reported that 74% recovered naturally.<sup>248</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>248</sup> the report provides no detailed information about treatment received by the recovered children.

The Illinois group reported results from another cohort of 81 pre-school children,<sup>249</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>223</sup> reported that, of the 51 children who stuttered at 3 years of age, 36 (71%) had recovered. 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>250</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 intervention during the pre-

school years. Fewer parents of children who recovered from stuttering—13%—reported that their child received treatment.

A large cohort report from the Twins Early Development Study<sup>143</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.

#### *Smaller cohorts*

A report of 23 stuttering pre-schoolers<sup>251</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>252</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.

Another report<sup>253</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>254</sup> involved a follow-up of 22 pre-schoolers who were diagnosed as stuttering 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. In light of 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 pre-school years, around two-thirds to three-quarters of children will recover naturally 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 to their children’s stuttering.

### **Prospective reports of natural recovery during the pre-school years**

#### *Why is this important?*

Lectures Six and Seven show that the merits of early intervention during the pre-school years have become apparent during recent decades. Hence it is now obvious that effective treatment needs to occur at some time during that period. 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 pre-school years is essential to consider during clinical decision making, as discussed during Lecture Seven.

#### *Three data sources*

Data from the Illinois Project<sup>248</sup> provide information about this matter. The mean age of the 84 children in that 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>229</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 stuttering treatment<sup>255</sup> also provided some indication of what the recovery rate shortly after onset might be. The trial had a control group of 25 pre-school



children who received no treatment for 9 months. Three of those children (12%) appeared to recover during that period. A limitation of this data set is that it was not designed as an epidemiological study and hence did not have enough children for any confident conclusions. However, its strength is that it was an exclusively clinical group of children.

### *Conclusions*

Based on those 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.

## **Is natural recovery predictable?**

### *Reported family history*

A review<sup>125</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 stuttering pre-schooler will recover. A study of 1,043 relatives of 66 children who stuttered<sup>256</sup> reported a genetic trend for persistence and recovery from stuttering. The large cohort Twins Early Development Study<sup>143</sup> mentioned earlier replicated that finding.

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

The ELVS report on 4-year-olds<sup>229</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>228</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 natural recovery. The report also linked maternal education to recovery, including a 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>250</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 not. The odds ratio was 7.1, with a 95% confidence interval of 1.3–37.9. That means, in short, that the result should be considered with caution, 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 treatment for stuttering.

### *The Purdue Stuttering Project*

A report from the Purdue Stuttering Project<sup>257</sup> involved 74 children who stuttered with a mean age of 57 months at the start of the study. Steep growth of syntactic development during yearly clinic visits during 3 years was reported to predict natural recovery at the end of that period. 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 a number of 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 were at the end of their pre-

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

school years at the time the study began. Another reservation is that the findings are of theoretical interest only, because it is not clinically viable to monitor children for 3 years for signs of natural recovery. It is also the case that the report does not indicate how many of the recovered children received treatment. But perhaps most importantly, children with mild stuttering were considered to have recovered from stuttering: children scoring 1 on a severity scale where 0 indicated *normal* and 7 = *very severe*. 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.

Those results are consistent with a report from the same cohort<sup>258</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. However, this report contained the limitation of other reports from this cohort that children with mild stuttering were considered recovered. Eight of 19 (42%) of the recovered group had received treatment.

A smaller scale report from the Purdue Stuttering Project,<sup>259</sup> reported predictors of natural recovery for 40 pre-schoolers who stuttered and 25 controls. The children were followed for a mean of 38 months until median ages 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 caveat for the other report of this cohort just discussed<sup>257</sup> applied in this case, with children having mild stuttering classified as recovered. An additional caveat about interpreting the findings was that 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 statistically unable to adjust for this potential confound. Perhaps surprisingly then, the authors concluded that “these results indicate that preschool articulation abilities should be part of a comprehensive assessment for determining risk of chronic stuttering” (p. 41).<sup>257</sup>

#### *The Illinois cohort*

The two prospective Illinois cohorts have contributed preliminary suggestions about the predictability of natural recovery. The first cohort<sup>260</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>232</sup> of 81 children provided similar suggestions, adding increased variability of jaw displacement and negative temperament as predictors of early stuttering persistence. However, these results were presented with the qualification that “results were not definitive” (p. 12).

Another preliminary finding emerged from first Illinois cohort about a connection between natural recovery and breastfeeding.<sup>261</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 odd ratio, making the finding difficult to interpret. Regardless, the finding can 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.

#### *A small cohort*

The small-cohort study mentioned earlier<sup>251</sup> for 23 stuttering pre-schoolers generated data about predictors of recovery 6 years later.<sup>262</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.

*So is natural recovery predictable?*

The short answer to the question of whether natural recovery is predictable arguably should be similar to the answer to the previous question of whether stuttering onset is predictable. In a statistical sense yes, but in a clinically useful sense not really. There is little convincing evidence that a clinically significant proportion of natural recovery can be accounted for by case variables. The available evidence is seriously compromised by the effects of treatment and questionable criteria for recovery from early stuttering. It seems that reported family history of recovery—which implicates genetic factors—is suggestive that a child with early stuttering will recover. The ELVS report—as yet unreplicated— suggests that boys have a much greater chance of recovery at 1 year post onset compared to girls. And there is a recurring connection in the literature between speech and language proficiency and the chance of natural recovery.

**SUMMARY**

Stuttering affects people throughout life from early childhood to old age, potentially as adversely as life threatening medical conditions. Stuttering affects quality of life across educational and occupational domains. Negative social stereotypes about the disorder are common, even among school teachers. The disorder commonly causes social anxiety, which is connected to situation avoidance. Genetics is involved with the disorder although not many details are known at present. Stuttering is a common disorder that is extremely prevalent during early childhood. A comprehensive database shows stuttering to be the equal third ranked of a range of developmental disorders. Its 4-year cumulative incidence could be one in nine children. Onset occurs early during the pre-school years unexpectedly, unpredictably, and often rapidly. Two-thirds to three-quarters of children will recover at some later time, however the probability of recovery during the first year after onset is low.

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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 disorder is to understand its nature and cause. So an obvious way causal theory influences daily clinical practice is when clinicians use it to explain what stuttering is and why clients or their children are affected by it.

##### **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 that 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.

Those researchers<sup>1</sup> showed that constructs of treatment credibility and expectancy were related to parent compliance with a health care treatment for their children. This issue has been found to be pertinent for one of the childhood treatments discussed later during these lectures:<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>

#### **AN EXAMPLE OF A CLINICALLY INFLUENTIAL CAUSAL THEORY**

##### **The Diagnosogenic Theory**

There are many early theories of stuttering that are now of historical rather than scientific interest, and they are overviewed in reference texts.<sup>4,5,6</sup> Examples include pyknolepsy theory, perseverative theory, approach-avoidance theory, the Orton-Travis Theory, two-factor theory, primary and secondary stuttering theory, and psychoanalytic theory. Johnson’s Diagnosogenic Theory is regarded as one of those theories now of historical interest.<sup>6,7</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>8</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 they in fact had normal disfluency. According to the theory, it was subsequently, when parents became anxious about these normal disfluencies and tried to make their children stop doing them, that stuttering developed. In Johnson’s words, stuttering began not in the mouth of the child but in the ear of the parent. The theory was formally proposed in 1942.<sup>9</sup>

Part of the extensive influence 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>10</sup> The advice offered there to parents was:

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



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)

The theory and the clinical advice that followed from the Diagnosogenic Theory attained widespread acceptance, and for decades no clinician, or anyone else it seems, would ever think of directly treating early stuttering by calling attention to it. The situation in the 1970s is portrayed here:<sup>6</sup>

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)

### The fall

The fate of most theories during the course of advancing knowledge in a field is for them to be wrong. They can't all be right,<sup>11</sup> and part of scientific development is the eventual realisation that a theory is wrong, and this is what happened with the Diagnosogenic Theory. Emerging research evidence during the 1970s strongly suggested that it was wrong. A prime example was the reports of verbal contingent stimulation of stuttering with pre-schoolers, described during Lecture One. The most famous of those was a 1972 publication<sup>12</sup> showing that two pre-school children reduced stuttering when attention was called to it, and a similar finding to the same effect was published some years later.<sup>13</sup> If the Diagnosogenic Theory was correct, then calling attention to children's stuttering would have worsened it, not improved it. The first public proclamation that the Diagnosogenic Theory was wrong was published in 1983,<sup>14</sup> but its influence lingered long after that.

There was much controversy and colour surrounding the theory. As mentioned during Lecture Two, Johnson conducted a dubious experiment during the 1930s that was not published<sup>15</sup> but which, decades later, was found to not support the theory<sup>16</sup> and which resulted in legal proceedings.

On the colourful side, the theory predicted that if a culture could be found with no word for stuttering or concept of what it was, there would be no stuttering in that culture. Johnson published a report in 1944<sup>17</sup> stating that the Bannock and Shoshone Indians of Idaho in the United States had neither any word for nor concept of stuttering, and none of them stuttered. Correspondence came to light in 1981<sup>18</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>19</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."

The Diagnosogenic Theory provides a telling illustration not only of how a causal theory can influence clinical practices, but how that influence can go awry. A recent report shows that, decades later, when the theory is obviously wrong, some clinicians still believe it to be true. A recent study of 37 speech-language pathologists and 70 speech-language pathology students<sup>20</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).

## TESTING CAUSAL THEORY OF STUTTERING

### Introduction

At first, it might seem that the notion of what causes a disorder, or anything to happen, is a simple matter. It is well worth reading a brief introduction to the philosophy of science that deals with causality, in the first two chapters of a landmark text.<sup>6</sup> 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, paradigms, laws and theories, models, and hypotheses. The discussion on pages 10 and 11 about what causes a bushfire gives some insight into the potential complexity of studying causality.

This is not to say that causality never is simple. The authors of that text mention single gene anomalies that cause human problems, such as cystic fibrosis or sickle cell anaemia. All those who have the genetic anomaly have the disorder and nobody has the disorder without the genetic anomaly. Or to say it another way, a single gene is necessary and sufficient to have the disorder.

That reference text<sup>6</sup> discusses four ways to evaluate a causal theory: testability, explanatory power, parsimony and heuristic value. The first two of these are now overviewed.

### Testability of a theory

The prime source of information used to evaluate theory is experimentation, which, to cut short a long story, is observations in contrived circumstances that make them more powerful. For example, imagine a theory of stuttering that the problem is in the larynx with vocal fold function. There have been such theories, with one in particular being prominent.<sup>21</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 (presumably) is no vocal fold function, and during standard speech, when there is vocal fold function. If the theory is correct and stuttering is a problem with vocal fold function, there should be no stuttering during lipped speech. In fact, such experiments have been done<sup>22,23</sup> and it is obvious that stuttering can occur during lipped speech.

The influential philosopher Carl Popper is credited with the axiom that experimentation does not prove a 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. A theory that constantly resists active attempts to disprove it attains increasing credibility. 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 from observational challenges.<sup>24</sup>)

### Explanatory power of a theory

The more that a theory can explain about its topic the more credible it is. Stuttering presents so many things that need to be explained by a theory, and a causal theory of stuttering needs to be evaluated in light of how well it explains them. The following are just some of the prominent research findings about stuttering that causal theories need to take account of in order to be credible.

#### *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 theory is that everyone who stutters does so in a different way, even though they obviously have the same disorder. 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 particular sounds and words. Stuttering occurs more commonly on content than function words. And a most obvious but commonly

overlooked fact for any causal theory to explain is that stuttering does not occur on every syllable spoken; it presents as an intermittent problem involving stuttering moments. It is even more challenging for causal theory to explain that language is not even necessary for stuttering to occur; stuttering can occur experimentally on non-words, where lexical processing is not necessary.<sup>25</sup>

### *Epidemiology*

As outlined during Lecture Two, stuttering begins during the pre-school years, 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?

### *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>26,27,28,29</sup> One of those reports<sup>29</sup> provided acoustic evidence of this occurring. So whatever causes stuttering is capable of affecting a non-speech activity.

### *Stuttering and manual tasks*

Although there have been findings to the contrary, there are research findings that signs of the disorder are to be found outside the speech mechanism. Examples include delayed manual reaction times for those who stutter<sup>30,31,32</sup> and finger movement tasks.<sup>33,34,35</sup> There have 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>36,37,38,39,40,41,42,43</sup> Such results have been independently replicated,<sup>44,45,46</sup> and interest in the topic seems to persist with a further replication.<sup>47</sup> Compared to controls, those who stutter have been shown to have more timing asynchrony when playing piano melodies.<sup>48</sup> All this is even more intriguing than findings about playing wind instruments and speaking nonsense words, because such tasks have nothing at all to do with speech. There is evidence, however, that the effect is not present with pre-school children who stutter,<sup>49</sup> raising the suggestion that the effect is connected with the effect rather than the cause of stuttering.

### *Stuttering severity is variable*

Stuttering severity is notoriously variable. It is likely to vary with differing audience sizes and types,<sup>50,51,52</sup> generally with more stuttering as audiences become larger. Stuttering severity varies also across different everyday situations.<sup>53</sup> It seems that there will be more stuttering when speaking to people than when speaking alone.<sup>52,54,55,56</sup> The latter study showed with a group a 60% stuttering reduction when alone compared to when an experimenter was present. Experiments that have involved repeated measures of participants in the same speaking situation have shown clinically significant stuttering variability in that same situation.<sup>57,58,59</sup> A study of six participants over five clinic visits spanning 2 weeks<sup>60</sup> showed that in two cases stuttering severity was four or five times greater on some visits than others.

Statistical process control charts are a method of studying variation, and that method has been applied to stuttering.<sup>61</sup> The stuttering severity of 10 adults was studied during the course of their speech during a single day. Results showed that all 10 showed predictable variation around their mean severity. However, five of the participants had stuttering severity that was unpredictable, and suggestive of an

“out of control system,” showing severity scores more than three standard deviations from their means during the day.

#### *Stuttering and genetics*

Any causal theory of stuttering needs to be able to explain, as outlined during the previous lecture, that genetics is obviously involved with stuttering. Although details are not fully known at present, inheritance of stuttering obviously is complex, with a number of genes involved.

#### *Brain structure and function*

Also as discussed during the previous lecture, any causal theory of stuttering needs to take account of research findings connecting stuttering and problems with brain structure and function. Those findings are suggestive of a genetically determined problem with myelination of white-matter tracts.

## MULTIFACTORIAL MODELS OF STUTTERING CAUSALITY

### **The fundamental proposition**

In short, multifactorial models state that stuttering is caused by the interaction of many factors to be found in the living environments of early childhood and within children themselves. There is nothing necessarily pathological about the factors involved. They just interact uniquely for each child to be responsible for stuttering. To say it precisely, these models specify nothing as necessary or sufficient for stuttering development.

### **The Demands and Capacities Model**

The best known multifactorial model is the Demands and Capacities Model, which, as its name implies, states 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 proposed: speech motor control, language development, social and emotional functioning, and cognitive development. In the words of its proponents, “there is no single etiology, but as many etiologies as there are stories of stuttering development” (p. 24).<sup>62</sup>

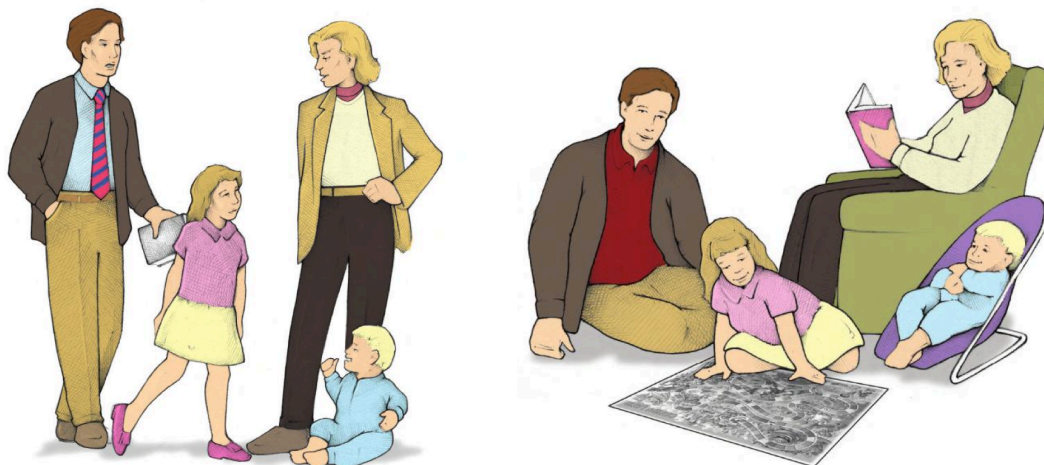
The prominence of the Demands and Capacities Model prompted an entire issue of the *Journal of Fluency Disorders* to be devoted to it in 2000. The model has been described at many sources,<sup>62,63,64,65,66,67</sup> with its first appearance in a 1987 textbook:<sup>68</sup>

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)

The model has been depicted graphically<sup>69</sup> as shown in these figures.<sup>‡</sup> The first shows a situation, on the left, where demands exceed capacity, and hence where stuttering occurs. The situation on the right shows a scenario when capacities exceed demands and stuttering does not occur.

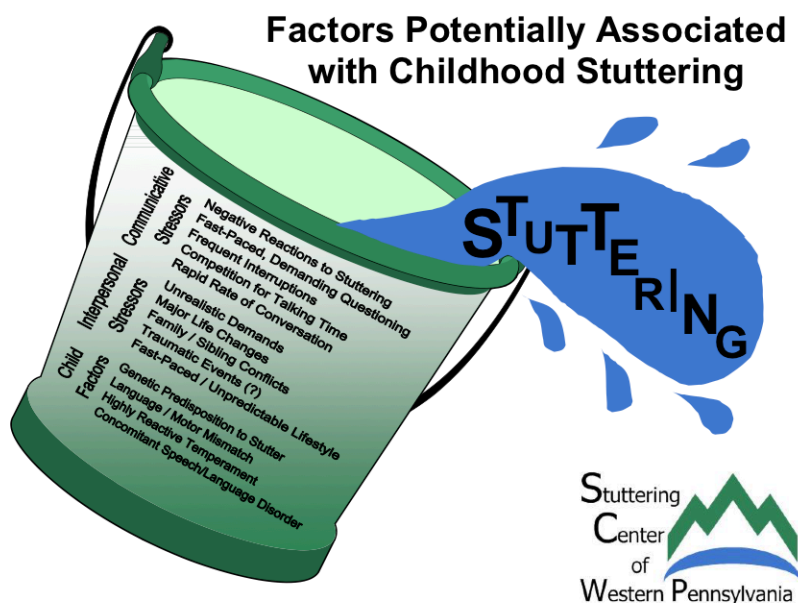
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<sup>‡</sup> Reproduced with permission: Guitar, B (2014), *Stuttering: An integrated approach to its nature and treatment* (4th ed), Baltimore, MD, Lippincott Williams & Wilkins.



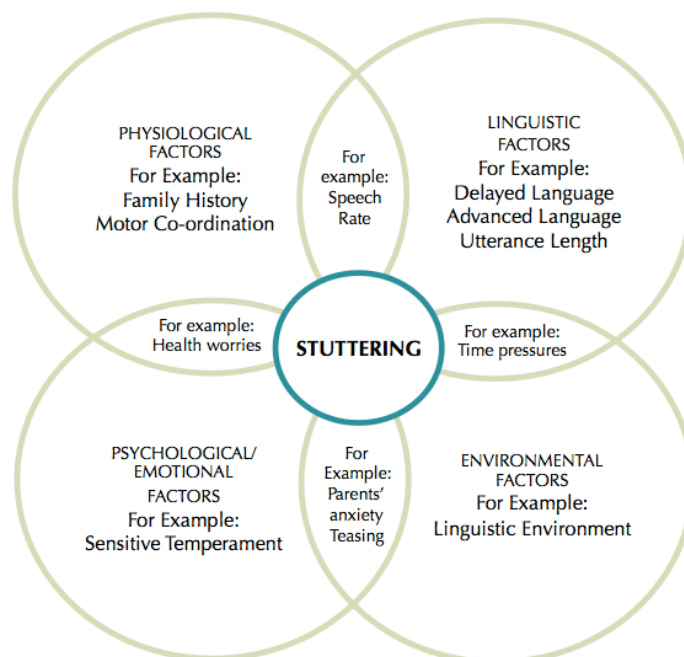
**Other multifactorial models**

There have been several variants of multifactorial causal models in addition to the Demands and Capacities Model. Two are used internationally as stuttering treatment models,<sup>70,71</sup> particularly in the United States. The next figure<sup>‡</sup> is a graphic from the Stuttering Center of Western Pennsylvania.<sup>71,72</sup> The conceptual similarity between it and the Demands and Capacities Model is apparent, as is the notion that nothing is necessary or sufficient for stuttering, as shown by the phrase “factors potentially associated with childhood stuttering.”



<sup>‡</sup> Reproduced with permission: the Stuttering Centre of Western Pennsylvania, © 2004.

The Michael Palin Centre in London proffers another variant of multifactorial models,<sup>73</sup> shown in the next figure.<sup>‡</sup> Again, the conceptual similarity with other multifactorial models is apparent. Another variant is known as the Dual Diathesis-Stressor Model,<sup>74</sup> which includes a temperamental proclivity component.



### Testability

Although extensively popular theoretically and clinically, the Demands and Capacities Model has been criticised many times,<sup>6,75,76,77,78,79,80</sup> and those criticisms imply criticisms of multifactorial models in general. Those criticisms reiterate the point that multifactorial models are 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>62</sup> It is logically impossible to disprove an indefinite number of causes.

### Explanatory power

These models do not score well in terms of explanatory power. An obvious problem for them is explaining the epidemiological fact that most stuttering appears during such a narrow age range during the pre-school years. Such models would suggest that it could begin at any time during childhood family life when the factors sufficient for stuttering converge, creating a situation where demands for fluency exceed the child's capacity to produce it. Also from an epidemiological perspective, it is problematic that the models specify that a cause of stuttering is located in the living environments of early childhood. How could it be, then, that stuttering persists throughout life when that early childhood environment no longer exists? It is logically essential that "all causal factors must be operating at every moment of stuttering" (p. 226).<sup>75</sup> For a related reason, multifactorial models do not explain stuttering variability across time and situations throughout adult life. However, they do explain why stuttering might vary during early childhood in different speaking situations; different situations

<sup>‡</sup> Adapted and reproduced with permission: The Michael Palin Centre, © 2014.

involve a different mix of demands and capacities. The table presents a suggested summary of the explanatory power of multifactorial models.

<i>MULTIFACTORIAL MODELS</i>	
<i>RESEARCH FINDING</i>	<i>EXPLANATORY POWER</i>
<i>Behavioural diversity</i>	<i>No</i>
<i>The influence of spoken language</i>	<i>No</i>
<i>Epidemiology</i>	<i>No</i>
<i>Conditions that reduce or eliminate stuttering</i>	<i>No</i>
<i>Stuttering and wind musical instruments</i>	<i>No</i>
<i>Stuttering and nonsense words</i>	<i>No</i>
<i>Stuttering and bimanual motor sequences</i>	<i>No</i>
<i>Stuttering severity is variable</i>	<i>No</i>
<i>Stuttering and genetics</i>	<i>No</i>
<i>Brain structure and function</i>	<i>No</i>

### **The future of multifactorial models**

Multifactorial models were first proposed two and a half decades ago. From a scientific perspective they have attracted much criticism. Such criticism is justifiable, considering that they are logically impossible to test and that their explanatory power is questionable. Rather than providing theoretical understanding of why stuttering develops during early childhood, they seem only to restate the problem; children begin to stutter because they are unable to do otherwise. Regardless, multifactorial models currently enjoy clinical popularity as a basis for techniques to control of early stuttering, and they have prompted laboratory studies exploring their clinical usefulness<sup>81,82,83,84,85</sup> As discussed during Lectures Six and Seven, there have been two clinical trials of such techniques.

Variants of multifactorial models seem not to have sustained much interest so far this century, although they have been described in a clinical context within two book chapters,<sup>73,86</sup> and they still feature as topics of presentations at international conferences about stuttering. One peer-reviewed scientific journal publication<sup>87</sup> restates an existing model<sup>70</sup> with accompanying explanation of how it is broadly consistent with some aspects of current knowledge about the disorder. Yet the publication seems to add nothing about the explanatory power of the model.

## **THE INTERHEMISPHERIC INTERFERENCE MODEL**

### **The fundamental proposition**

It appears that the first formal proposal of this model, implicating the supplementary motor area, occurred in 1987.<sup>88</sup> The Interhemispheric Interference 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>89</sup> but departs from it by specifying that those who stutter have normal 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” (p. 125–126).<sup>90</sup>

The developer of this model, William Webster, has relied on logic derived from the research findings about bimanual sequence tasks discussed earlier:<sup>91</sup>

the neural systems underlying such sequential movement control overlap those involved in speech and orofacial movements. Accordingly, anomalies in sequential finger-tapping in stutterers may suggest something about the nature of the “aberrant interhemispheric relations” hypothesized by Orton and Travis. (p. 11)

This prompted the proposal that:<sup>36</sup>

Although sequential response mechanisms may be lateralized normally in stutterers, the repetitive sequential finger tapping error data suggest that these mechanisms may nonetheless be unusually susceptible to interference. (p. 818)

### Testability

The model is certainly consistent with many brain imaging findings of unusual hemispheric speech processing with those who stutter. A recent review<sup>199</sup> overviews that literature. Yet a large-scale study of the planum temporale<sup>†</sup> was not consistent with the model.<sup>92</sup> It refuted earlier findings<sup>93,94</sup> of differences in symmetry between stuttering and control participants for that anatomical structure. It has been argued<sup>6</sup> that the model is difficult to refute experimentally because neither of its two brain components are operationally defined: the inefficient supplementary motor area and the over-reactive process of hemispheric activation. However, the model developers reported that it was verified with an experiment<sup>88</sup> where stuttering and control participants performed a finger-tapping task with a concurrent task using the other hand. The stuttering participants had more interference from the concurrent task than the controls.

Another experiment, though, caused a problem for the model by showing that the same result occurred with a bimanual writing task: writing with both hands concurrently.<sup>95</sup> The results were consistent with a cognitive problem rather than a physical problem with concurrent left and right handed activity. Webster described the problem in a later publication:

it is unlikely that the interference with sequencing mechanisms in stutterers is strictly an interhemispheric phenomenon as was suggested by the studies of bimanual co-ordination ... it unlikely that the origin of that interference is limited to callosal influences.<sup>97</sup> (p. 12)<sup>†</sup>

The model received its most sophisticated experimental test using a combination of finger tapping and bimanual crank turning tasks, and two judgement tasks involving the left and right visual fields.<sup>90</sup> The experiment was designed to determine whether the model could explain natural recovery in terms of speech motor control maturation, specifically in the supplementary motor area. Participants were those who had recovered from stuttering, those with persistent stuttering, and controls. Consistent with previous findings, recovered stuttering participants and controls performed better than stuttering participants with the bimanual tasks. However, the stuttering and recovered stuttering participants performed equally poorly on the visual tasks, suggesting that the latter group retained residual interhemispheric problems.

### Explanatory power

The model certainly can explain the manual sequencing anomalies that have been found in those who stutter. However, its explanatory power is weakened by not only the experiment with bimanual writing tasks described earlier, but by problems with it incorporating other research findings about

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<sup>†</sup> The planum temporale is an anatomical structure associated with language function, and it typically is asymmetrical between the two hemispheres.

<sup>†</sup> Collosal refers to the corpus collosum, which is a large white matter fibre structure connecting the two hemispheres of the brain.



writing tasks.<sup>96,97</sup> Additionally, explaining the influence of spoken language and stuttering variability seems problematic. That aside, the model does have considerable explanatory power. It is able to explain at least some of early epidemiology with the natural recovery study.<sup>90</sup> It might also explain the narrow range of stuttering onset in terms of early language development exposing the underlying hemispheric problem at that developmental stage. It can explain the fluency enhancing conditions because they could simplify speech motor activity to compensate for a problem with interhemispheric speech processing. Stuttering with wind instruments, nonsense words and bimanual tasks can be explained by the model, because it does not specify that the brain problems are speech specific.

*INTERHEMISPHERIC INTERFERENCE MODEL*

<i>RESEARCH FINDING</i>	<i>EXPLANATORY POWER</i>
<i>Behavioural diversity</i>	<i>No</i>
<i>The influence of spoken language</i>	<i>No</i>
<i>Epidemiology</i>	<i>Yes</i>
<i>Conditions that reduce or eliminate stuttering</i>	<i>Yes</i>
<i>Stuttering and wind musical instruments</i>	<i>Yes</i>
<i>Stuttering and nonsense words</i>	<i>Yes</i>
<i>Stuttering and bimanual motor sequences</i>	<i>Yes</i>
<i>Stuttering severity is variable</i>	<i>No</i>
<i>Stuttering and genetics</i>	<i>Yes</i>
<i>Brain structure and function</i>	<i>Yes</i>

### The future of the Interhemispheric Interference Model

Webster acknowledges in several publications that the broad notion underlying his theory has a long history. It dates back to the early years of the last century and Lee Edward Travis<sup>†</sup> who proposed the Orton-Travis theory of cerebral dominance,<sup>89</sup> the origins of which are apparent in a 1925 report about dyslexia.<sup>98</sup> In 1978 Travis recounted tests of the Orton-Travis theory that were presented in a 1931 textbook,<sup>99</sup> long before the arrival of scientific journals in the discipline.<sup>100</sup>

When I published the cerebral dominance theory of stuttering in 1931, I presented in its support three laboratory findings: (1) reductions of the patellar tendon reflex latency, (2) reductions in the amplitude of tremors from extended right forefingers, and (3) profound alterations in the alternating phasic movements (opening and closing) of both hands, all during tonic stuttering blocks ... (p. 278)

From all accounts, those were ground breaking research methods for the field.<sup>101</sup> In a 1978 publication,<sup>100</sup> Travis outlined how the theory was able to explain a series of research findings in the 1960s and 1970s. 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>102</sup>

That historical background could prompt speculation about the future of this idea about stuttering causality. It appears to be an intrinsic part of thought and research in the discipline about the nature

<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.

and cause of stuttering. It might be interpreted as an encouraging sign that, for more than 80 years now, the Interhemispheric Interference Model has resisted definitive experimental disproof and its future is auspicious. In fact, as discussed during the previous lecture, there is now evidence of anomalies in the corpus callosum—the white matter structure connecting brain hemispheres—among children who stutter. The current iteration of the model has had, overall, encouraging empirical verification and reasonable explanatory power. That might be interpreted as a sign that such a model, in some future iteration, may eventually be accepted as a causal explanation for the disorder.

So far, there has been little experimental interest in this theoretical explanation for stuttering apart from its developers. However, a recent report<sup>103</sup> used magnetoencephalography to study lateralisation of brain function with stuttering and control pre-schoolers during picture naming. No differences were reported for stuttering and control groups. Arguably, however, this result is not interpretable without evidence using similar research methods for older age groups, showing that they do have a lateralisation difference. It is the case, though, that magnetoencephalography has found interhemispheric differences with adults who stutter using other methods.<sup>104,105,106</sup>

## THE COVERT REPAIR HYPOTHESIS

### The fundamental proposition

#### *Drawing on Levelt's model*

This theory draws on Levelt's well known model of speech production,<sup>107,108</sup> and another model of phonological coding.<sup>109</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 selection of a word in abstract form (lemma) and its grammatical encoding. Finally, a "mental syllabry" is accessed<sup>110</sup> and the word becomes a set of syllables ready for articulation.

#### *Phonological coding errors*

The central proposition with the Covert Repair Hypothesis is that those who stutter have phonological coding errors in this process of preparation for articulation, and that stuttering moments are covert attempts by the speaker to correct those errors before speech execution of the faulty plan.<sup>111,112</sup> Those who stutter have more errors than those who do not, and consequently they need to correct those errors more. Those corrections occur before the articulatory sequence occurs, and this leads to repeated movements and fixed postures during speech.

#### *A continuum*

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 need to make more corrections. In effect, the hypothesis proposes that stuttering and normal disfluency are on either ends of a continuum. As mentioned during Lecture One, this is known as the Continuity Hypothesis.<sup>113</sup>

### Testability

#### *Confirmations*

The developers of the Covert Repair Hypothesis, the Dutch researchers Postma and Kolk, have presented support for the hypothesis using research methods involving speech errors of stuttering participants.<sup>114,115</sup> Other researchers have provided supportive data for the hypothesis,<sup>116</sup> reporting that 5 and 6 year old stuttering children had significantly lower "phonological memory" than control children, as measured with a non-word repetition task. Those same researchers provided further supportive data,<sup>117</sup> reporting that a group of stuttering children with a mean age 5 years 7 months had inferior performance to control children on sound blending and elision tasks. (Elision is removing a phoneme from one word to create a new word.) Other researchers have reported that 11 year old children who stutter are slower with a phoneme monitoring task,<sup>118</sup> and less accurate than controls with non-word repetition and phoneme elision tasks.<sup>119</sup> That research group reported that adults who

stuttered were slower than controls in repeating non-word phoneme sequences.<sup>120</sup> With adults, researchers<sup>121,122</sup> have reported that a stuttering group showed unusual responses to phoneme monitoring tasks compared to a control group.

#### *Reported falsifications*

However there have been several empirical reports by that claim to falsify the hypothesis. One finding<sup>123</sup> with nine boys who stuttered with normal phonology, and nine boys who stuttered with disordered phonology, was that neither group showed more self-repair behaviours than the other. Another report<sup>124</sup> with 12 stuttering boys of mean age 55 months tested the prediction of the hypothesis that higher articulatory rate would cause more stuttering, but it was not found to be so. Another report<sup>125</sup> of 32 adult stuttering participants and 32 controls found that the former made more errors with tongue twisters, which was consistent the hypothesis. However, the number of errors made did not correlate with any stuttering severity scores, which would not be predicted by the hypothesis.

Another challenge to the hypothesis occurred<sup>126</sup> with a study of 12 stuttering and 12 control children ages 7–12 reciting a list of nonsense words. No significant differences in errors could be found. Another report<sup>127</sup> was of a man who stuttered but produced phonological errors when he did so only on stuttering moments that were “part-word repetitions.” The authors claimed this as a challenge the hypothesis. Two publications produced lip electromyographic data with an experimental reaction time paradigm, and argued that the results were inconsistent with a motor planning problem.<sup>128,129</sup> The most recent empirical report<sup>130</sup> compared speed of phonological encoding with stuttering adults and controls and concluded that the data did not support the Covert Repair Hypothesis.

#### **Explanatory power**

The Covert Repair Hypothesis has certainly prompted interest in the current literature, with many examples of researchers using it to broadly frame or interpret their research. The hypothesis indeed does have strong explanatory power, as argued by its developers.<sup>111</sup> It can explain the fundamental epidemiological fact that stuttering onset occurs some time shortly after the beginning of speech development; phonological complexity gradually increases with language development to a point where the pathological aspects of the problem become apparent. It explains the fluency enhancing conditions that might involve speech rate reduction; speech rate reduction would reduce the inherent problem for those who stutter. The hypothesis can explain the repeated movements and fixed postures of stuttering quite well. For example, if an error is detected at the last sound of a syllable, then repetition of the initial sound and vowel that precedes it will occur until the correct sound is ready (for example, do-do-do-dog). The theory developers present detailed explanation of various stuttering types according to the theory<sup>112</sup> (Tables 1 and 2). However, the theory, like others, is silent about superfluous behaviours. The hypothesis can explain the anomalies of brain structure and function associated with stuttering, in terms of them being fundamentally responsible for phonological coding errors.

The hypothesis does, however, have some serious shortcomings with explanatory power. Its developers acknowledge<sup>111</sup> that the hypothesis does not explain the occurrence of natural recovery. Also, it does not explain why stuttering varies within people and across time and situations.

There are several research findings that are inconsistent with the hypothesis. It has been pointed out,<sup>123</sup> for example, that it is at odds with a finding<sup>131</sup> that speech rate and response latencies—the time taken for a verbal response during conversation—did not differ between stuttering and control pre-school children. The hypothesis would predict the opposite.

The hypothesis suggests that the problem of stuttering is constrained to phonology. As such, it is troubled by the findings that those who stutter perform less well with activities that are phonology independent: playing wind instruments and bimanual tasks. The theory has shortcomings with explaining how language can influence stuttering.

#### *COVERT REPAIR HYPOTHESIS*

RESEARCH FINDING	EXPLANATORY POWER
<i>Behavioural diversity</i>	Yes
<i>The influence of spoken language</i>	No
<i>Epidemiology</i>	No
<i>Conditions that reduce or eliminate stuttering</i>	Yes
<i>Stuttering and wind musical instruments</i>	No
<i>Stuttering and nonsense words</i>	Yes
<i>Stuttering and bimanual motor sequences</i>	No
<i>Stuttering severity is variable</i>	No
<i>Stuttering and genetics</i>	No
<i>Brain structure and function</i>	No

### The future of the Covert Repair Hypothesis

The Covert Repair Hypothesis is eminently testable and has generated much research. However, so many failures to verify the theory, from different researchers using different methods, casts some doubt on its longevity. A review of the pertinent literature in one report<sup>130</sup> attributed such results to the range of stuttering severity and types of stuttering involved, and the different research methods used. Another viewpoint would be that if the covert repair hypothesis is true, then phonological encoding problems with those who stutter should occur lawfully and be detected easily with all samples of stuttering participants across a range of research methods. Another reviewer of the theory concluded “the covert repair of errors of phonological encoding cannot account for all instances of disfluency associated with stuttering” (p. 25).<sup>132</sup>

If the Covert Repair Hypothesis should, during the next few years, head toward being one of those theories of historical rather than current interest to the discipline, then that will be worthwhile progress. Expedient abandonment of a theory based on empirical research is welcome progress for a discipline, and clinicians who are ultimately guided by theory about stuttering would be well informed by such a development.

## THE EXPLAN THEORY

### The fundamental proposition

*Also drawing on Levelt’s model*

This theory has in common with the Covert Repair Hypothesis that it draws on Levelt’s speech processing model to specify a cause of the stuttering problem.

*A delayed motor plan*

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>133,134,135</sup> The impetus for its development seems to be that stuttering generally tends to occur more often on content than function words. The authors propose a hypothesis that the “stuttering of function words is caused by unavailability of instructions for the following content word” (p. 1020).<sup>133</sup> They propose also that, compared to function words,<sup>133</sup>

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)

It appears that the first formal statements of the theory occurred some years later,<sup>136,137</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>138</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 because they are more difficult to plan than function words. The theory suggests that<sup>139</sup>

whole-word repetitions (and also pauses) are ways of stalling motorically (repeatedly executing a previously generated program) on material prior to other material that is difficult to plan ... whereas prolongations part-word repetitions and word breaks reflect planning problems (the repetition, prolongation and hesitation within words signify that the plan was not right or was only partly prepared). Prolongations, part-word repetitions and word breaks are referred to as advancing to indicate the speaker has moved forward prematurely in the speech stream and to contrast with what happens in stalling. (p. 56)

So, stuttering occurs when the speaker either 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.

#### *A continuum*

As with the Covert Repair Hypothesis, the theory incorporates the Continuity Hypothesis, linking the normal disfluency of early childhood to stuttering development. This argument is stated clearly in one publication,<sup>135</sup> and draws on the notion that “young speakers, whether they are diagnosed as stutterers or not, would exhibit similar nonfluencies” (p. 346).<sup>135</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 chronic stuttering. Early onset stuttering is simple repetition of function words, to delay things, because the content word is not ready. Chronic 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**

It might be arguable that research thought to confirm and falsify the Covert Repair Hypothesis, as discussed earlier, could have the same impact on the EXPLAN theory. Regardless, the EXPLAN development team reported “very few dyfluencies” (p. 345)<sup>135</sup> for children or adults on function words that occurred after content words, which was interpreted as supporting the theory. That paper also argued it was consistent with the theory that for all age groups “dysfluency ... occurred predominantly on either the function word preceding the content word or on the content word itself, but not both” (p. 345).<sup>135</sup>

Recently an independent group directly tested EXPLAN theory<sup>140</sup> with an argument that, during early stuttering, it predicts that the phonetic complexity of the second word of an utterance will predict whether a stuttering occurs on the first word of an utterance. The authors found that for 14 three-year-olds that was not the case.

#### **Explanatory power**

The leader of the development team for this theory points out its strengths in explanatory power.<sup>138</sup> It offers an explanation for the influence of spoken language on stuttering. It explains the intermittent nature of stuttering; why it does not occur on every syllable. The reason is that delays in motor planning occur according to the difficulty of what is being planned. It explains the prominence of stuttering on function words during early stuttering and the switch to prominence of content words

during chronic stuttering. As is the case with the Covert Repair Hypothesis, which also deals with the speech process, it explains the early time of life during which stuttering onset occurs, and it does credibly explain different types of stuttering moments. The theory, like others, is silent about superfluous behaviours. As with the Covert Repair Hypothesis, it does explain the fluency inducing conditions in terms of reduced speech rate, and the origins of the problem in brain structure and function.

Being a theory that deals with specific interruption to the process of speech production, it shares shortcomings with the Covert Repair Hypothesis. It does not explain why natural recovery occurs. Nor does it explain why stuttering onset can be sudden or gradual. Being a speech process theory, it shares with the Covert Repair Hypothesis that it cannot explain findings about playing wind instruments and bimanual tasks. It also seems to be a fatal problem for the theory that stuttering can occur experimentally on non-words, where lexical processing is not necessary. It shares with the Covert Repair Hypothesis a shortcoming with explaining why stuttering varies within people and across time and situations.

#### EXPLAN THEORY

RESEARCH FINDING	EXPLANATORY POWER
<i>Behavioural diversity</i>	Yes
<i>The influence of spoken language</i>	Yes
<i>Epidemiology</i>	No
<i>Conditions that reduce or eliminate stuttering</i>	Yes
<i>Stuttering and wind musical instruments</i>	No
<i>Stuttering and nonsense words</i>	No
<i>Stuttering and bimanual motor sequences</i>	No
<i>Stuttering severity is variable</i>	No
<i>Stuttering and genetics</i>	Yes
<i>Brain structure and function</i>	Yes

#### The future of the EXPLAN Theory

It is difficult to offer any projection about the future of the explain theory because formal statements of it only emerged this century, barely a decade ago. The theory is testable and perhaps it will resist disproof with experimentation during coming years by researchers independent of its development. At present, though, its weakness seems to be its limited explanatory power.

## THE P&A MODEL

### Background

Packman and Attanasio developed the P&A Model,<sup>75,141</sup> advancing earlier thinking outlined in previous publications.<sup>142,143</sup> This model is multifactorial, but, for two reasons, not in the sense of the multifactorial models discussed earlier. First, the models discussed earlier specify no factors as necessary and sufficient for stuttering, but the P&A Model specifies factors that are necessary and sufficient for stuttering. Second, the P&A Model seeks to explain the occurrence of *individual stuttering moments*, not the development of the disorder. As such, the logic underpinning the development of the P&A model is that “all causal factors must be operating at every moment of stuttering” (p. 226).<sup>75</sup> A

model needs to explain how all causal factors are operating to cause every stuttering moment that occurs during life.

The developers of the P&A Model acknowledge 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>144,145,146</sup> They credit Wingate with the notion that prosody, of which syllabic stress is a part, is somehow disturbed with stuttering.<sup>147,148,149</sup> Wingate recognised that the effect of rhythmic stimulation involved changes to stress, and specifically that rhythmic speech reduces stress contrasts.

### The fundamental proposition

There are three factors in the P&A Model, as follows.

#### *Impaired neural processing for spoken language*

The model assumes a central nervous system problem of some kind that gives some children an inherently unstable speech motor system. That problem manifests as a deficit in neural processing of spoken language that makes it prone to perturbation. In the model, the neural speech processing problem 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.

#### *Triggers for stuttering moments*

The underlying neural speech processing problem and triggers for stuttering moments are, together, necessary and sufficient for stuttering moments to occur. Moments of stuttering are triggered by certain features of spoken language. These are the linguistic variables that increase the motor task demands placed on the already unstable speech system. They are the varying of stress or emphasis from each syllable to the next, and linguistic complexity. That pushes those who stutter beyond what their unstable speech system can deal with, triggering stuttering moments.

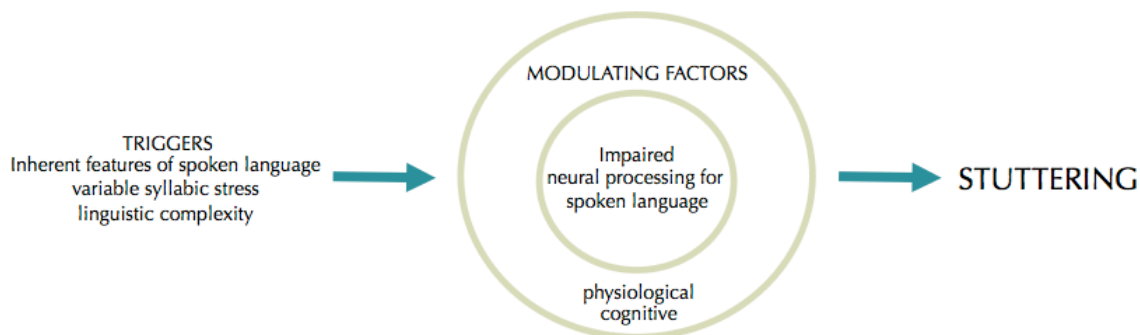
This idea draws on the watershed time during early language development when children begin to produce linguistic stress contrasts. So, for example, they will say “*dad-da*,” emphasising the first syllable, instead of “*dad-da*.” That may not seem much of a difference, but it is a leap of speech motor control. Subsequent to initial triggering of stuttering moments by early attempts to produce linguistic stress contrasts, maladaptive responses by children to struggle with the problem causes continued stuttering development.

#### *Modulating factors*

According to the P&A Model, the threshold above which moments of stuttering are triggered is modulated, differently for each individual, according to level of physical arousal at the time. Anxiety is a prominent cause of physiological arousal, which may lower the threshold for stuttering moments to occur and may be associated with more of them occurring. Cognitive factors might also lower the threshold for stuttering moments to occur. The model draws on evidence that stuttering increases with the physiological arousal presumably associated with increased audience size,<sup>150,151,152</sup> The model also draws on evidence that stuttering increases when a competing linguistic activity diverts attention away from speaking.<sup>153,154</sup>

The P&A Model is illustrated in the figure.<sup>‡</sup>

<sup>‡</sup> 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.



### Testability

The model is a recent development, so it is too soon to expect a critical test of it. One such test would be to scan the brains of genetically at-risk infants prior to stuttering onset. Because the model posits a central nervous system anomaly is a necessary condition for stuttering, no child who develops stuttering should be without such an anomaly prior to stuttering onset. Another prediction of the model would be that variability of vowel duration increases more during a short period just prior to stuttering onset than it does during an adjacent earlier period.

The model would be challenged if stuttering did not decrease under experimental conditions that reduced linguistic stress contrasts. Such an experiment should be possible because acoustic correlates of linguistic stress are well known, including syllable intensity, fundamental frequency and duration. The theory developers 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>143</sup>

### Explanatory power

The P&A Model was designed specifically to explain research findings about stuttering, so not surprisingly, it does so well, as shown in the table. Its original development was intended to explain findings that vowel duration variability decreases with treatments that incorporate a fluency enhancing condition.<sup>155,156</sup> Indeed, the model can explain those treatments in terms of reduced vowel duration variability that compensates for speech motor system instability by reducing linguistic stress during speech.

The model can explain the prominence of repeated movements at stuttering onset. They are the child’s response to the problem by attempting 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. The model incorporates current knowledge about brain structure and function to explain onset during the years of language development. If myelination is involved in the neural speech processing problem, the P&A Model can explain natural recovery for some children and a lifetime of stuttering for others. The posited underlying problem with neural speech motor processing can explain differences in stuttering severity across situations and within individuals. The severity of the underlying neural processing problem would influence the baseline severity of stuttering across individuals, and the modulating effects of physiological and cognitive factors would explain idiosyncratic stuttering differences in differing speaking situations and times.



*P&A MODEL*

<i>RESEARCH FINDING</i>	<i>EXPLANATORY POWER</i>
<i>Behavioural diversity</i>	Yes
<i>The influence of spoken language</i>	Yes
<i>Epidemiology</i>	Yes
<i>Conditions that reduce or eliminate stuttering</i>	Yes
<i>Stuttering and wind musical instruments</i>	Yes
<i>Stuttering and nonsense words</i>	Yes
<i>Stuttering and bimanual motor sequences</i>	Yes
<i>Stuttering is variable</i>	Yes
<i>Stuttering and genetics</i>	Yes
<i>Brain structure and function</i>	Yes

The model explains much of the influence of spoken language, because stuttering on initial sounds and initial words of utterances is associated with linguistic stress. Stuttering is more likely to occur on linguistically complex than simple utterances. The model is consistent with evidence that linguistically complex utterances contribute to instability of speech movements.<sup>157</sup>

One limitation of the explanatory power of the P&A Model is that it would suggest, overall for those who stutter, a systematic and measurable relation between physiological arousal and stuttering rate. However, experiments about that matter have failed to show such a systematic relation. Such a relation has not been found with stuttering contingent electric shock,<sup>158</sup> with challenging interviews compared to supportive ones,<sup>159</sup> with feared sounds,<sup>160</sup> or with standard reading and conversation tasks.<sup>161</sup> Additionally, there seems to be no relation between self reported anxiety and either subjective or objective measures of stuttering severity.<sup>162</sup> Another issue with the explanatory power of the model is shared by all others reviewed during this lecture: the developers have not offered an explanation why around a third of children begin to stutter suddenly, during the course of a single day, and how verbal response contingent stimulation can control stuttering so well during early childhood.

**EPILOGUE**

Many theories developed during the last century have not attracted any interest during this century according to publications in peer-reviewed journals or published conference proceedings. Arguably, a theory that did not is potentially destined to join those with an historical place in the discipline. These include the Sensory-Motor Modelling Theory,<sup>163</sup> the Neuroscience Model,<sup>164</sup> the Anticipatory Struggle Hypothesis,<sup>165</sup> the Two-Factor Theory,<sup>166</sup> the Neuropsycholinguistic Theory,<sup>167</sup> and the Suprasegmental Sentence Plan Alignment Model.<sup>168</sup> Those theories are overviewed in several reference texts.<sup>4,5,6</sup> Should any such theories generate future interest they will feature in subsequent iterations of these lectures. Also, in the event that a theory is proposed during this century and generates interest, it will be included in these lectures. For example, a theory proposed this century<sup>169</sup> might well generate such interest because it comprehensively explains how speech treatments and various speaking conditions (see Lecture One) can reduce or eliminate stuttering.

In any event, the coming and going of so many causal theories about stuttering during the past century reflects its status as a puzzling disorder, arguably among the most puzzling that has affected humans. It is difficult to say whether the progress in understanding stuttering causality is slower than might be expected for such a disorder, or whether progress might be considered satisfactory. Still, there is an

overarching impediment to clarifying the cause of stuttering that seems to recur during the research overviewed during this lecture. The contents of Lectures One, Two and Ten show stuttering to be associated with diverse effects on people spanning speech motor function, psychology, phonetics, language, and speech acoustics. The disorder has been associated with anomalies across all those domains, but the direction of any causality has yet to be established. In other words, those anomalies could be associated with the cause of the disorder, or its effects, or an interaction between the two. That is a conundrum for research intended to explore the cause of stuttering.

## **SUMMARY**

The cause of the disorder is a necessary topic for discussion with clients and parents, and causal theory guides treatment development in the long term. The viability of a theory includes its testability and explanatory power. Five causal theories of stuttering have attracted interest during this century: multifactorial models, the Interhemispheric Interference Model, the Covert Repair Hypothesis, the EXPLAN Theory, and the P&A Model. On balance, no causal theory has yet resisted experimental disproof sufficient times or over a sufficient period to warrant any confidence. At present, though, it seems reasonable to say that stuttering appears to be associated somehow with a problem of neural speech processing, although details are far from clear. That broad idea, in various iterations, has resisted disproof since the early years of the last century.

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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 nonbehavioural. Much of the nonbehavioural impact of stuttering will be related to anxiety, and can be measured with procedures 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.

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

##### **Communicating with clients**

Clinical measures establish a common language between clinician and clients or parent 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 in order 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 intended to achieve. Clinical stuttering measures convey to clients or their parents what those intended achievements are. The formal description of this process uses terms such as setting of treatment targets or 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 standard for the clinician and client to determine the treatment goals together.

Using measurement to document treatment goals and whether they have been met is part of treatment accountability. Stuttering treatment has to be funded, either through government or private sources. Those who provide that funding have an interest in knowing 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 for 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 toward treatment goals. If progress toward those goals is not satisfactory, clinical measures will assist with documenting and exploring why that is so, so that the problem can be fixed.

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<sup>†</sup> Thanks to Sue O’Brian for guidance with this material.

### 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 clinic for more treatment, perhaps several times, after their original treatment. As discussed during 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 is known to occur.<sup>2</sup>

Clinical measurement can be used to monitor clients' 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 designed to prevent relapse is referred to as maintenance, and is an indispensable part of any stuttering treatment.

### Keeping track of daily stuttering severity changes

As noted during 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 more stuttering typical of 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 authority figures. It is essential for clinicians to use clinical measurement to know about and keep track of such day to day variation 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.

## PERCENTAGE SYLLABLES STUTTERED (%SS)

### Overview

This is a measure of the percentage of spoken syllables that are stuttered, commonly abbreviated to %SS. 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 refers to moments during speech that are clearly stuttering and not normal disfluency.

Percentage syllables stuttered is based on syllables spoken, the syllable being a fundamental unit of speech production.<sup>3</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 pre-school years the ratio is much lower at 1.15 according to one source.<sup>4</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. There is evidence that a %SS score during a 10-minute everyday conversation is representative of stuttering severity during that entire day.<sup>5</sup>

There is information from the latter study<sup>5</sup> that 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

“l,” and two attempts to say it, does not change the fact that, for the purposes of calculating %SS, it was just one stuttered syllable.

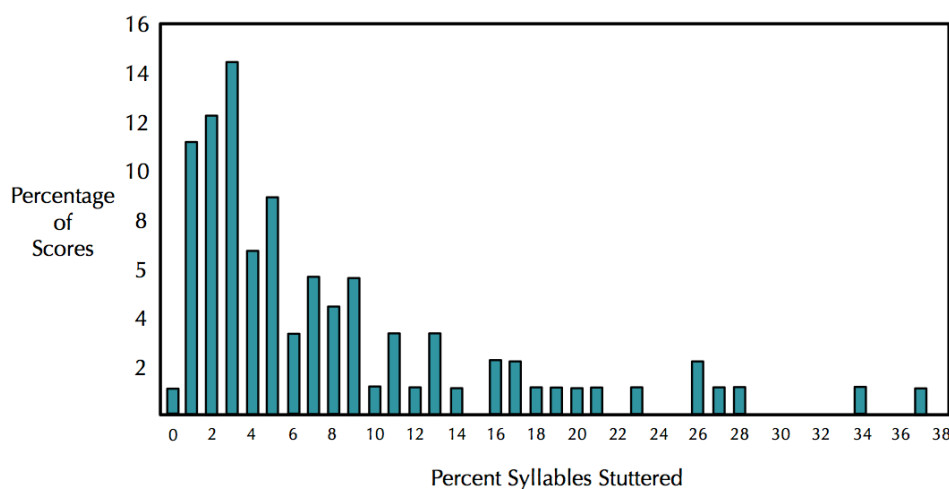
Less commonly, percentage words stuttered is used, which is a similar measure but calculated by counting every word spoken, not every syllable spoken. That measure is now used rarely for research and clinical practice.

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

With a clinical measure it is convenient if the population values are normally distributed, so the mean score falls in the middle of a normal distribution, with half the scores above the mean and half below the mean. Then, it is known that around two-thirds—68.2% to be exact—of cases are within one standard deviation either side of the mean. So that helps to interpret extreme scores in terms of how far from the mean they are.

The distribution of %SS scores is not normal. There are more mild cases than severe cases.<sup>6,7,8</sup> Information from the latter report<sup>8</sup> about adults is reproduced in the diagram.<sup>‡</sup> 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 pre-school children,<sup>9</sup> although %SS scores generally seem to be lower at that time of life.<sup>6</sup> In the figure, 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.<sup>†</sup>



<sup>‡</sup> Adapted and reproduced with permission: O’Brian, 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.

<sup>†</sup> 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.

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 have been discussed in detail.<sup>10</sup>

### Equipment for percentage syllables stuttered measurement

During clinical practice, %SS is typically measured during a conversation with the client, or during conversation with a child or while watching a conversation between a child and parent. A two-button counting device is used for measuring %SS, such as the one in the photograph. 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>11,12</sup>

Considerable training is required to learn to use such equipment, but such training is readily available.<sup>11,12</sup> A disadvantage of using a smartphone application for measuring %SS is that the buttons are not mechanical. This may make it difficult to maintain eye contact with the client while measuring %SS online, as the clinician is doing in the photograph.



### Limitations of percentage syllables stuttered

#### *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 %SS, this is not a clinical option. Consequently, clinical use of %SS is normally constrained to measurement in the clinic by the clinician. An option to obtain %SS measures 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 in to the clinic. Such recordings can be made easily with smartphones. Although it is time consuming to measure %SS from such recordings, in some clinical contexts the effort would be justified.

#### *Reliability*

Reproducibility refers to how well a measure gives the same score when it is used several times in the same way.<sup>13</sup> Reliability is a more commonly used term for this concept.

Percentage syllables stuttered is a notoriously unreliable measure. The first report about this was in 1940,<sup>14</sup> showing that 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>15</sup> One paper<sup>16</sup> lists 32 research reports to that effect. Among other reliability problems, those research reports show that if one observer gives a certain %SS score there is no guarantee that another observer will give the same score or a similar score. With research, this is known as a problem with inter-judge agreement, sometimes called inter-observer agreement. Poor inter-judge agreement has been reported many times with %SS measures.<sup>17,18,19,20</sup>

In the most notorious of those studies,<sup>17</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 much 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>21</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>22</sup> was adapted for stuttering.<sup>23,24,25</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>26</sup> showed that this method did not solve the reliability problems with stuttering-count measures. Also, it 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>27,28</sup> The latter study<sup>28</sup> reported also that it did not help to slow down speech samples while counting syllables and stuttering moments. A recent report about training procedures for stuttering counts<sup>29</sup> was more encouraging, however the training by no means solved the reliability problems with stuttering counts.

## SEVERITY RATING (SR) SCALES

### Overview

#### *Differing numbers of scale divisions*

Severity rating (SR) scales are a different type of clinical measure to %SS. Severity ratings are perceptual measures, where an observer listens to a sample of stuttered speech and uses the 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, and 10-point scales are commonly used. Often, but not always, some or all of the scale divisions have labels telling the user what they represent.

#### *Commonly used clinical severity rating scales*

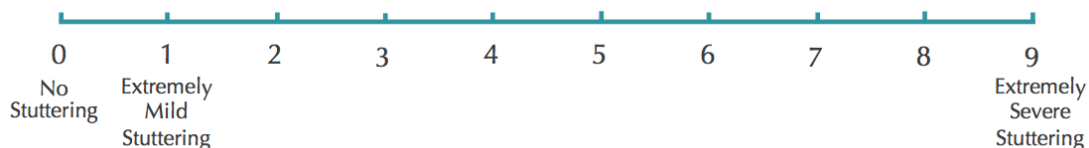
Severity rating scales used in research often have seven scale divisions. Examples are used in the Illinois Early Childhood Stuttering Project (see Lecture Two). One version is 0 = *normal disfluency*, 1 = *very mild stuttering*, and 7 = *very severe*,<sup>30</sup> and another version is 0 = *normal speech*, 1 = *very mild stuttering*, and 7 = *very severe stuttering*.<sup>31</sup> Clinical scales in common use have nine or 10 scale divisions. A scale commonly used during treatment of pre-school children (see Lecture Six) is 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 9 = *extremely severe stuttering*. A scale commonly used for older children, adolescents and adults (see Lectures Eight and Nine) is 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 8 = *extremely severe stuttering*. Scores are commonly written in clinical files as SR 1, SR 2, SR 3, and so on.

#### *A behavioural measure*

Severity ratings have in common with %SS that they are intended as behavioural measures of stuttering severity. When clients score their own SRs their scores may be inclined to some extent to reflect their nonbehavioural experiences with the disorder, notably speech anxiety. So it is important to instruct clients not to allow such factors to influence their SR scores; their speech related anxiety can be measured using procedures outlined during Lecture Eleven.

#### *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:



During clinical practice, clients can use an Internet-based platform, such as Google Forms,<sup>32</sup> to record their severity rating scores, as shown in the diagram. Client severity ratings populate a spreadsheet with the date and time that each severity rating was entered. If needed, a continuous graph of client severity ratings can be established with a regression line of best fit to detect any trends.

**Jamie's Daily Stuttering Severity**

0 = No Stuttering

1 = Extremely Mild Stuttering

2

3

4

5

6

7

8

9 = Extremely Severe Stuttering

**SUBMIT**

### Equal interval ordinal scales

These 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 psychophysics. That topic has been covered with specific reference to ordinal scaling of stuttering measurements.<sup>33</sup>

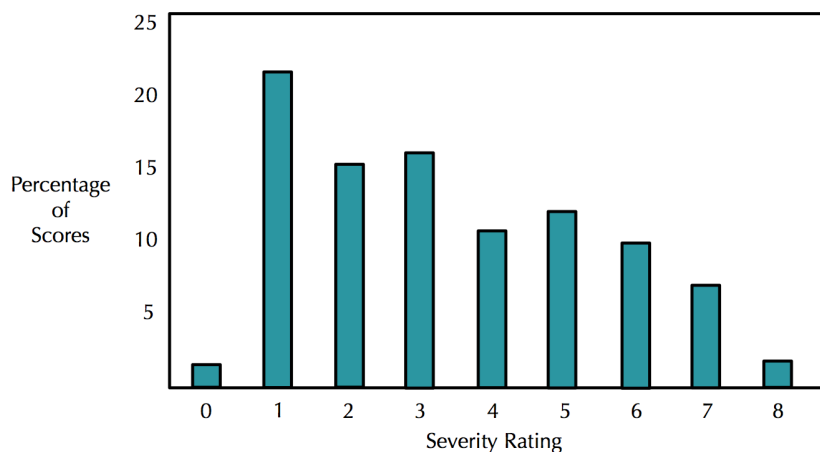
### Severity rating scores are not normally distributed

As is the case for %SS scores, SR scores are not normally distributed. The graph<sup>‡</sup> shows clinician SRs on a 9-point scale using data from a report mentioned earlier with 90 adult stuttering participants.<sup>8</sup> The scale is 0 = no stuttering, 1 = extremely mild stuttering, and 8 = extremely severe stuttering. It shows mean SRs rounded to the nearest whole scale value. Cases that scored up to a mean SR 0.4 were rounded down to SR 0. Although these scores are not plainly normally distributed, they are certainly more normal looking than the previous graph of %SS.<sup>†</sup> The median score is SR 3.0.

<sup>‡</sup> Adapted and reproduced with permission: O’Brian, 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.

<sup>†</sup> The paper used a 1–9 scale, but the graph converts the data to a 0–8 scale.





### Reliability of severity ratings

Logically, there is more chance of attaining adequate reliability with SR than with %SS, simply because SR scales have much fewer potential scores. By 2011 there were 11 research reports about SR reliability for stuttering<sup>16</sup> (see Table 1, p. 1287). Those reports show that while SR scales are not altogether free of problems, they are probably more reliable than %SS scores. There is evidence that shows SR scores to be more reliable than %SS in a clinical context where there is a need to detect changes over time with individual clients.<sup>16</sup>

For children, there is some evidence that, with little training, clinicians and lay listeners agree when using such scales,<sup>34</sup> and that parents of children who stutter have close rating agreement with clinicians.<sup>35,36</sup> One report<sup>37</sup> used the 10-point SR scale described earlier with 3–6 year olds speaking 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. However, one report<sup>38</sup> showed that clinicians do not use the scale reliably when adult clients speak an unfamiliar language of Mandarin.

Ideally, for inter-judge agreement, clients and peers would all give the same speech sample the exact same SR. 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.

### A severity rating scoring guide†

After listening to a client for whatever period seems reasonable as a valid speech sample, these three questions can be used to guide the assigning of a SR.

*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 normal 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*.

† Thanks to Sue O'Brian for this material.

*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 that 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.

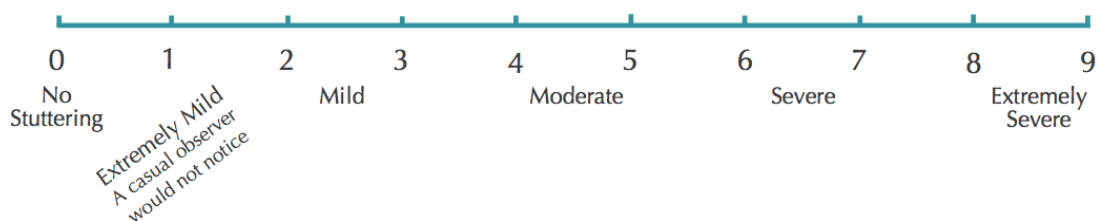
*How much does it affect communication?*

As discussed during Lecture Two, 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 disfiguring stuttering may be because of superfluous behaviours. In cases where superfluous behaviours are particularly disfiguring, a clinician may feel that communication is particularly affected.

*Was it mild, moderate, or severe?*

It is useful to think of four categories to describe how the stuttering affects communication: *mild*, *moderate*, *severe*, or *extremely severe*. *Mild* would be SR 1–3, *moderate* would be SR 4–5, *severe* would be SR 6–7, and *extremely severe* would be SR 8 (or 8–9 in the case of a 10-point scale). The categories *extremely mild*, *mild*, *moderate*, *severe* and *extremely severe* are useful, incidentally, as informal descriptions of client stuttering severity. A report might read, for example, “I assessed this 37-year old man today who presented with extremely mild stuttering.”

The diagram is a summary of the guide for a final decision about a SR score.



### 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 and mentoring from a senior colleague. There are no generally available training methods to show inexperienced clinicians what a group of experienced clinicians believe are representative SR scores for the clinical population.

Clinical knowledge about the severity of a 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 his 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*

In contrast to %SS, a compelling advantage of SRs is that they are simple and require no equipment, which means they cost nothing. Additionally, it seems that extensive training is not needed to learn to use them,<sup>34,35,36</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 without the need for detailed translation.<sup>39</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 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 considerable clinical flexibility. For example, a clinician may ask a client or parent to record a “typical SR” and “worst SR” during a defined period during a day, or for an entire day.

### *Valid*

It seems that SRs are more valid than %SS because they take account of all behavioural features of stuttering rather than just a count of stuttering moments. There is some evidence that clinician SRs take account of some relevant information about severity that %SS does not, which is discussed shortly.

### *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 are able to use SRs to measure their children’s stuttering at any time of the day when they are together.

## THE RELATION BETWEEN %SS AND SR

### 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>8</sup> This means that they can be used interchangeably with some confidence, but with two reservations. In that study,<sup>8</sup> what prevented a higher correlation were several cases where the %SS score did not at all correspond 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>40</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 %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>41</sup> (Clinical measurement during clinical trials is discussed during the next lecture.)

### Repeated movements and fixed postures

Repeated movements generally are not as disfiguring 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 those stuttering moments with repeated movements are particularly severe stuttering.

Conversely, consider a speech sample that has a quite low %SS score because there are quite few stuttering moments, but those few stuttering moments are fixed postures and they are particularly disfiguring and time consuming. Such a sample might score a low %SS but a higher SR because the disfiguring 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<sup>‡</sup> shows the comparative percentile ranks for the two measures for an adult caseload.<sup>8</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

### The relation between %SS and SR during treatment

Appendix One to this lecture is an example of a clinical file showing the use of %SS and SR measures during treatment of a child for 12 weeks. The clinical file illustrates the association between the two measures. The clinician has made a %SS measure in the clinic each week as well as recording parent SRs for each day of the week prior to each clinic visit.

The child scored SR 0 consistently for the last few weeks of the file record, with only the occasional SR 1, and the clinician %SS scores were virtually zero for that period. 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. The clinician %SS scores verify that result.

## SYLLABLES PER MINUTE (SPM)

Sometimes a clinical measure of speech rate, most commonly syllables per minute, is associated with %SS. Devices that measure %SS normally 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 many 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>42</sup>

## SPEECH NATURALNESS (NAT) MEASUREMENT

### Why measure speech naturalness?

Speech restructuring treatments (see Lecture One) are clinically useful for reducing stuttering but may not produce speech that sounds completely natural. This has been known to be clinically problematic for a long time.<sup>43</sup> Speech restructuring treatments involve a trade-off between speech that has no stuttering moments or a few stuttering moments, and speech that sounds natural. So a measure of speech naturalness is necessary for such treatments to measure how natural clients sound and to guide

<sup>‡</sup> Adapted and reproduced with permission: O'Brian, 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.

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

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>44,45,46,47</sup> For research purposes, it has been shown mathematically that:<sup>48</sup>

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)

There is evidence<sup>49</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.

## STUTTERING-LIKE DISFLUENCIES

Stuttering-Like Disfluencies is a measure that is used throughout the research publications from the Illinois Early Childhood Stuttering Project (see Lecture Two) and has been used also by some other researchers as a measure of stuttering severity:<sup>50</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 normal disfluency.<sup>51,52,53,54</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 words, implying that nonstuttering children show speech behaviours that can be referred to as “stuttering-like.” The potential problem arising from this paradoxical terminology is illustrated by a publication<sup>55</sup> that used the measure and had this wording in its title: “... frequency of stuttering in young children who do and do not stutter” (p. 2133).

## THE STUTTERING SEVERITY INSTRUMENT (SSI-4)

The Stuttering Severity Instrument<sup>56</sup> is now in its fourth edition, often abbreviated to SSI-4. It 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, duration of the three longest stuttering moments, verbal and nonverbal superfluous behaviours, and speech naturalness. The speech naturalness scale is the one just described. 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. There are normative data for 72 pre-school children, 139 school-age children and 60 adults. In order to use this measure the forms and manual need to be purchased from the publisher.

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 clinic visit.

There have been several reports questioning the reliability of this measurement instrument, which are overviewed in a more recent report that again questioned its reliability.<sup>57</sup> There is a report<sup>58</sup> to show

that the SSI-3 (the previous version to SSI-4) provides no additional information than can be obtained from a SR scale. Considering that, and considering that it involves expense to purchase and clinical time to complete, 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 OVERALL ASSESSMENT OF THE SPEAKER'S EXPERIENCE OF STUTTERING (OASES)**

Chronic stuttering affects quality of life and can be measured with the Overall Assessment of the Speaker's Experience of Stuttering,<sup>59</sup> commonly known as the OASES. It is designed to reflect the World Health Organization's International Classification of Functioning, Disability, and Health.<sup>60</sup> Each of the OASES questions requires a response on a 5-point scale, with higher scores reflecting more adverse quality of life impact. There are OASES Australian normative data to supplement North American normative data.<sup>61</sup>

The OASES is a questionnaire with four categories of questions. These are 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 quite recently<sup>62</sup> and is starting to appear regularly in publications. To use this measure, the forms and manual need to be purchased from the publisher.

The OASES would be a useful part of a clinician's assessment tools for documenting client quality of life before and after treatment. For that 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>61,62</sup> and preliminary results for OASES-S and OASES-T are encouraging.<sup>59</sup> The OASES-S has been translated into Dutch (OASES-S-D)<sup>63</sup> and the OASES-A has been translated into Japanese (OASES-A-J)<sup>64</sup> and Hebrew,<sup>65</sup> with data showing it to be reliable and valid in those languages.

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

The Wright and Ayre Stuttering Self-Rating Profile (WASSP) is another health related quality of life measure that also was designed to reflect the World Health Organization's classification system.<sup>66,67,68</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 as a clinical and research measure, with intended clinical application for assessment and 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>68</sup>

The WASSP developers report that the test has adequate reliability and validity.<sup>68</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 often in the United Kingdom where it was developed, but availability elsewhere seems currently to be limited.<sup>68</sup>

### **A SIMPLE SPEECH SATISFACTION SCALE**

Often, clinical reports measure client speech satisfaction with a simple scale, such as a nine-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 young 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 the disorder that impact on the client.

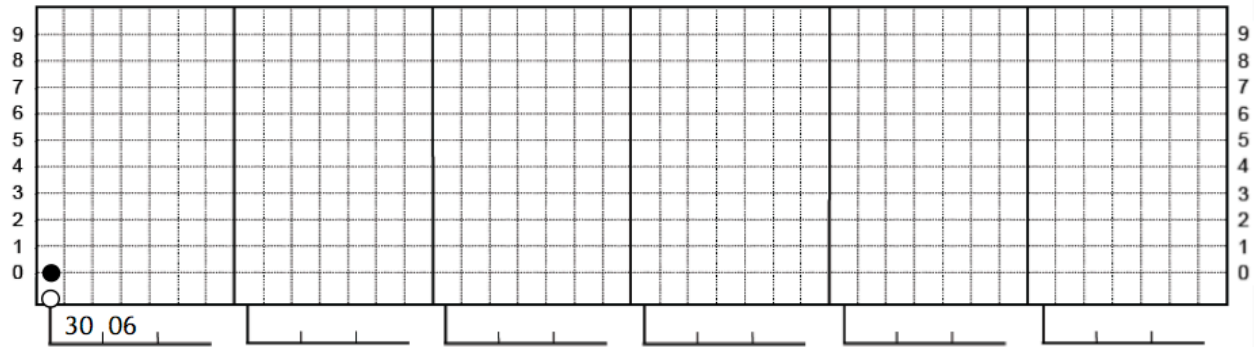
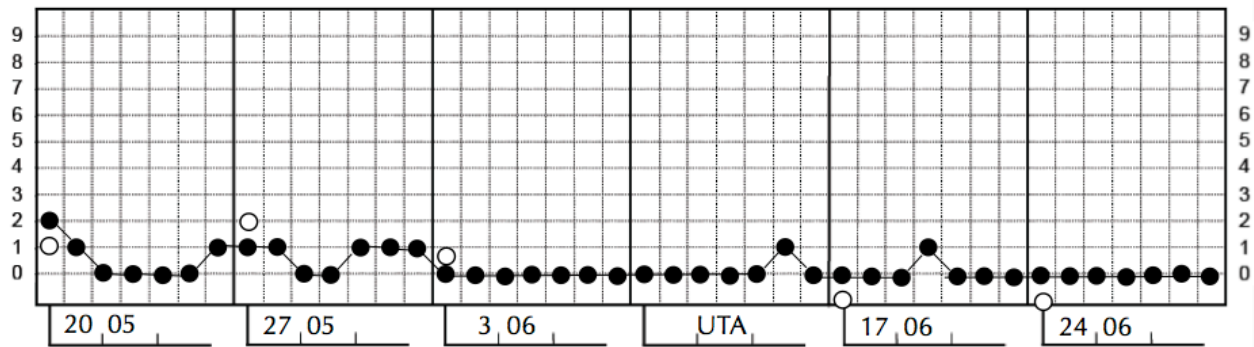
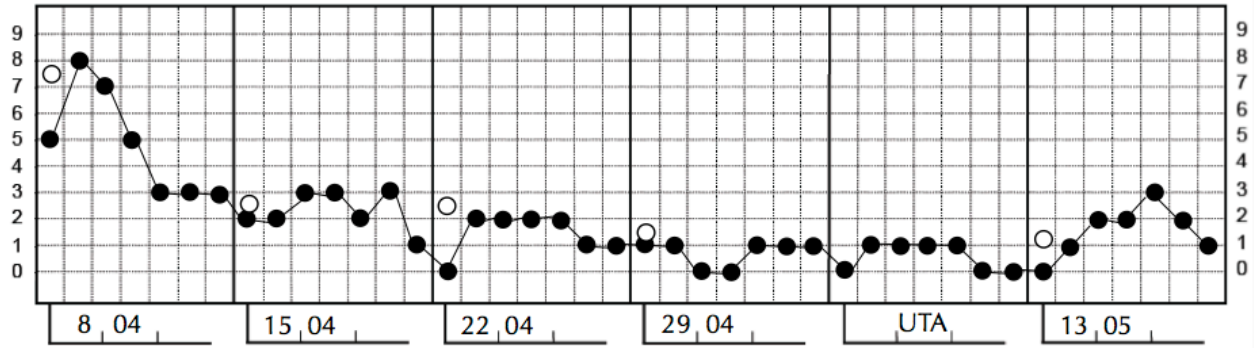
A more complicated version has been suggested <sup>69</sup> where clients make a judgment about their speech using a 10-point scale where 1 = *very bad* and 10 = *excellent*, with intermediate points on the scale labelled as *bad*, *very strongly insufficient*, *strongly insufficient*, *insufficient*, *sufficient*, *more than sufficient*, *good*, and *very good*.

## **SUMMARY**

Clinical measurement is essential to assess clients and communicate with them about their stuttering. It is also essential to state treatment goals, to assess progress toward them, and to manage the maintenance of those treatment goals. Stuttering severity can be measured most conveniently with %SS and SR. Severity rating measures have clinical advantages related to their simplicity and validity, and their covert use with clients when needed. Generally, %SS and SR measures seem to measure the same dimensions of stuttering severity, with some important caveats. Speech naturalness is an essential measure for treatments that involve a trade off between stuttering control and natural sounding speech. There are options available for measuring health related quality of life for clients before and after treatment. Clinical measurement procedures are summarised in Appendix Two.

**APPENDIX ONE**  
**%SS AND SR MEASURES DURING CLINICAL MANAGEMENT**  
**OF A PRE-SCHOOL CHILD**

Closed circles are parent SR scores for each day. Open circles are clinician %SS scores during each clinic visit. (UTA = unable to attend.)





## APPENDIX TWO

### SPEECH AND QUALITY OF LIFE MEASURES FOR STUTTERING

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<i>SPEECH</i>		
<i>Percentage syllables stuttered</i>	<i>%SS</i>	<i>A stutter-count measure of the proportion of spoken syllables that contains an unambiguous stuttering moment.</i>
<i>Severity rating</i>	<i>SR</i>	<i>A perceptual measure of stuttering severity using an ordinal scale.</i>
<i>The Stuttering Severity Instrument</i>	<i>SSI-4</i>	<i>A more detailed and time consuming measure of stuttering severity than either %SS or SR.</i>
<i>Syllables per minute</i>	<i>SPM</i>	<i>A measure of speech rate.</i>
<i>Speech naturalness</i>	<i>NAT</i>	<i>A perceptual measure of how natural speech sounds using an ordinal scale.</i>
<i>QUALITY OF LIFE</i>		
<i>Overall Assessment of the Speaker's Experience of Stuttering</i>	<i>OASES</i>	<i>A health related quality of life 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>
<i>Wright and Ayre Stuttering Self-Rating Profile</i>	<i>WASSP</i>	<i>A health related quality of life measure for adults, adolescents and school-age children involving domains of stuttering behaviours, thoughts about stuttering, feelings about stuttering, avoidance due to stuttering, and disadvantage 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, or evidence-based medicine as it is sometimes known, 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:<sup>2</sup>

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)

Another more recent definition explicitly mentions the mathematics involved with generating research evidence. Much research that is clinically useful—but not all of it—involves numbers of some kind derived mathematically.<sup>3</sup>

Evidence based medicine is the use of mathematical estimates of the risk of benefits and harm, derived from high-quality research on population samples, to inform clinical decision-making in the diagnosis, investigation or management of individual patients. (p. 1)

A comprehensive video overview is available,<sup>4</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 by far the largest professional speech pathology association in the world, and arguably the most influential. In 2005 it proclaimed:<sup>5</sup>

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. (p. 3)

#### **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:<sup>2</sup>

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)

##### **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 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 matter:<sup>6</sup>

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)

Some examples of knowledge you need for clinical practice that fall outside the domain of science are the benefits of empathy and emotional support for clients, listening skills, and hope and belief that intervention will help them. Those 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>7</sup> makes a point well 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>8</sup> Evidence-based practice has been applied to health rehabilitation generally<sup>9</sup> and specifically to speech-language pathology<sup>10</sup> and stuttering treatment.<sup>11</sup> An issue of the *Journal of Fluency Disorders* was devoted to the topic of evidence-based practice and stuttering.<sup>12,13,14,15,16</sup>

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

In order to be fully informed, clinical judgements need to be "moderated by patient circumstances and preferences" (p. 737).<sup>17</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. Evidence-based practice decisions may be different in that case to where parents are living together and the same 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 the entire speaking day.

At the most basic level, the clinician needs to determine why clients have come to the clinic. This idea of complaint-centred treatment is certainly not new for stuttering.<sup>18,19</sup> Broadly speaking, the problems that cause someone who stutters to present to a clinic will relate to either behavioural or nonbehavioural issues. Either there will be some need to control stuttering, or a need to deal with a nonbehavioural feature of the disorder, or a combination the two. Most likely, nonbehavioural issues will involve speech related social anxiety in some way, as discussed during Lectures Ten and Eleven.

Information pertinent to this matter was presented for the clinical experiences of 71 adult members of the National Stuttering Association, which is a United States self-help group for stuttering.<sup>20</sup>

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

Behavioural treatments to deal with stuttering were the most commonly reported intervention received, but 49% of respondents reported receiving treatment that “involved reducing the fear of stuttering or of speaking situations” (p. 120).<sup>20</sup> Fifty-three per cent reported a combination of behavioural and nonbehavioural approaches, and 25% reported nonbehavioural treatment “with little emphasis on speech” (p. 120).<sup>20</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 “their therapist did not seek information from them in the decision-making process” (p. 122) reported dissatisfaction with the treatment process.

With pre-school children it is virtually certain 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 discussed during Lecture Ten, from the school-age years through adolescence to adulthood, it seems that the likelihood increases of social anxiety becoming a clinically pertinent issue.

### **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 informs stuttering treatment: basic research, treatment process research, and treatment outcome research.

#### *Basic Research*

Basic research deals with the nature and the cause of the disorder. An example from Lecture One is research showing the possibility that a stuttering pre-school child will have another speech or language disorder. That information will influence your assessment procedures, and will need to be taken account of during clinical intervention if there is a stuttering comorbidity.

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 about when to begin early intervention, as discussed during Lecture Seven.

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 theories. In deciding whether to intervene with a stuttering pre-schooler using a treatment based on the Demands and Capacities Model, a clinician may wish to form a view about whether that theory is substantiated by basic research.

#### *Treatment process research*

Treatment process research deals with how treatment functions and factors that can affect how it functions. An example from the previous lecture is that speech restructuring treatments involve a trade off between stuttering control and natural sounding speech. That information guides 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 the previous lecture. That research may influence a clinician’s treatment process in various ways. For example, it might prompt a clinician to constantly calibrate %SS scores against a community reference, such as the training sources mentioned during Lecture Four.<sup>21,22</sup>

#### *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>23</sup> They provide credible 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 for consideration with clients. Or a clinical trial may show a treatment to be of limited or no of 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 do the treatment and evaluate its effects. This can be done with regular application of simple clinical measures outlined during Lecture Four, such as %SS and SR. 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 those clinical measures had not shown the change that 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 creditable to warrant consideration. A defensible rule of thumb here is whether the research has been published in a peer-reviewed journal that is listed in a creditable data base such as PubMed or the Web of Science. Other publication sources are sometimes referred to as the “grey literature,” and include, in addition to non-peer reviewed journals, student theses, books, book chapters, and internal institutional reports.

That being 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>24</sup> This has prompted a strong caution that “some ... published articles belong in the bin, and should certainly not be used to inform practice” (p. 31).<sup>3</sup> 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.

### **Hierarchies of evidence**

Subsequent to a decision about whether research is sufficiently creditable 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. Prominent examples are the University of Oxford Centre for Evidence-Based Medicine,<sup>25</sup> the National Health and Medical Research Council of the Australian Government,<sup>26</sup> and the Cochrane Consumer Network.<sup>27</sup>

A common theme appearing in all of them 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>28</sup> is a well established and trusted online source of systematic reviews.

Apart from a systematic review, those hierarchical classifications<sup>25,26,27</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. Those 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>25</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 scientific research to guide treatment has been described as assertion-based practice.<sup>29</sup> Experts, many of them charismatic, commonly proclaim the merits of certain treatments when there is no creditable research evidence to support their claims. The media often endorses such claims because of their charismatic nature.

### Detailed methodological critique

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. That judgement needs to be informed by detailed and rigorous scientific knowledge. For example, a clinician may devalue the importance of a report on the grounds that the authors used a misleading statistical analysis procedure. There are many sources of guidance for how such detailed critiques of scientific publications might be conducted.<sup>3,30</sup> Such texts often contain checklists for evaluating scientific publications, which include detailed items such as “if the statistical tests in the paper are obscure, why have the authors chosen to use them?” and “were outliers analysed with both common sense and appropriate statistical adjustments?” (p. 223).<sup>3</sup>

There is a website available that provides methodological critiques of treatment reports in speech-language pathology,<sup>31</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>32</sup> which clinicians can use to critically appraise stuttering treatment research evidence. The authors reported that inexperienced judges are able to use it reliably. The checklist was based on research standards that have historically been endorsed by leading scholars and researchers in the field.

That proposed checklist<sup>32</sup> has some controversial features. Its authors acknowledge that the randomised controlled trial is the gold standard for health research. However, they argue that it should not necessarily be placed at the top of the hierarchy of evidence for use by clinicians for evidence-based practice with stuttering because “the vast majority of stuttering treatment research uses other designs” (p. 127).<sup>32</sup> Another controversial feature of this checklist is that it does not necessarily require data to be collected by the standard “blinded” method, where the observer who collects the data does not know anything about the research or whether speech samples are pre-treatment or post-treatment. As a precursor to their checklist, the authors argue that stuttering treatment research ideally should have the following five fundamental methodological credentials.

- (1) A randomised design or a single-subject time series experimental design.
- (2) Data collected by a blinded observer or an unblinded observer who has agreement with a second blinded observer.
- (3) Treatment outcome measures at pre-treatment, during treatment, and at post-treatment.
- (4) Outcome measures are collected in the clinic and outside the clinic.
- (5) When a report shows reduced stuttering, speech rate and speech naturalness are shown to be normal.

The single-subject time series experimental design referred to in the first point involves many types of research designs,<sup>33</sup> which are sometimes called N=1 or N-of-1 trials.<sup>34,35</sup> They are recommended for use in situations which include rare disorders where it is difficult to obtain sufficient participant numbers for traditional clinical trials. However, this is not the situation with stuttering, which is quite a common disorder, as discussed during Lecture Two. A suggested summary of current views about N-of-1 trials in speech-language pathology<sup>36</sup> is that, to be compelling, they should incorporate “replication in at least five studies showing similar treatment effects with at least 20 patients and involvement of at least three research teams in at least three institutions” (p. 244).

## 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<sup>37</sup>

... any research study that prospectively assigns human participants or groups of humans to one or more health-related interventions to evaluate the effects on health outcomes.

The National Institutes of Health, which funds health research in the United States, defines a clinical trial this way:<sup>38</sup>

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

The major Australian health funding bodies and Australian Universities involved with health research define it this way:<sup>39</sup>

A clinical trial is a form of human research designed to find out the effects of an intervention, 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)

For the present purposes, the following definition of a clinical trial is suggested because it was designed specifically with reference to stuttering treatment, and incorporated reasonable consensus from within the field:<sup>23</sup>

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 pretreatment 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)

Although that definition of a clinical trial is used throughout these lectures, it is an arbitrary matter. Reviews of the evidence for stuttering treatment efficacy present differing views about what should be regarded as a clinical trial of stuttering treatment.<sup>40,46</sup> The first of those papers, for example, presents a far more liberal view of the matter than just described, including reports that contain “any outcome relating to a positive effect on ... communication or ... social and emotional wellbeing” (p. 678)<sup>40</sup> without regard to follow-up or whether speech measures were collected beyond the clinic. That being said, some details about the present working definition<sup>23</sup> are as follows.

## Clinical trial standards

### *Prospective methods*

Using the definition just outlined,<sup>23</sup> and in fact with 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>41</sup> and clinical follow-up of such cases.<sup>42</sup> That is not to say that retrospective reports of stuttering treatment outcome are not useful publications to consider during evidence-based practice. On 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. 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 is capable of producing evidence that a treatment can stop the developmental course of stuttering.<sup>43</sup> However, clinical experiments do not contribute information about the outcome of the entire treatment.

*Beyond-clinic speech measures*

It is a generally accepted rule about clinical behaviour change that it should be measured beyond the clinic to be sure that it has really occurred.<sup>44</sup> That is because of what is known as discriminated learning, which refers to 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 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>45,46,47,48,49,50</sup> The most common 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 remained 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, with clinical trials of drugs there are normally more participants 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>51</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 credentials, a Phase III clinical trial is far more compelling than a Phase I clinical trial.

**The CONSORT statement**

Standards for an acceptable clinical trial are specified in the Consolidated Standards of Reporting Trials (CONSORT) Statement.<sup>52</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 that receives the treatment. Measures are made pre-treatment and then post-treatment. Because of the few participants that 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. 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.

### *The importance of randomisation*

There is a well-known effect where non-randomised trials overestimate what the true effect size is.<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 pre-school children, 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 a trial can be. 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 quite 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 discussed shortly. It is necessary when beginning a Phase III trial to have some idea of the effect size, because it is used mathematically to determine how many participants are needed for such a trial.

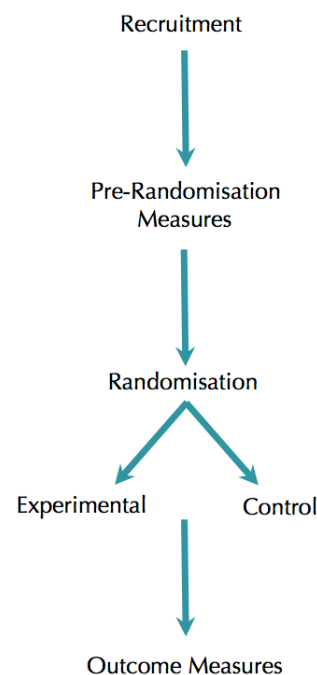
## Phase III clinical trials

### *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 do. A comprehensive guide to Phase III clinical trials of stuttering is available,<sup>54</sup> written specifically for a speech-language pathology audience. Salient points from that guide are summarised in the following.

### *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 not referred to as pre-treatment measures because, in a randomised controlled trial involving a no-treatment group, half the participants in fact receive no treatment.



*Randomisation*

Then, an independent person randomises the participants. Ideally, that independent person is a biostatistician, or is 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 to 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 compared to a treatment given to participants by telepractice using webcam. Sometimes with clinical trials that compare two treatments there is also a no-treatment control arm.

*Primary outcomes*

In a clinical trial, outcome measures are fundamental to how the outcome of the trial is judged. There is a rule specified in the CONSORT Statement that a randomised trial can have no more than two primary outcomes. The reason given for this is that, from mathematical and logical viewpoints, more than two outcomes makes it difficult to interpret the results of the trial.

*Secondary outcomes*

Although two is the maximum number of primary outcomes for a randomised trial, there can be several secondary outcomes. Secondary outcomes are measures that are used to complement the primary outcomes as measures of interest. For example, a randomised trial of a treatment to control stuttering may have %SS as the primary outcome, and as secondary outcomes might have 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 to 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 is generally considered to be favourable. There are related indices 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.

However, that 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. That could well be a statistically significant difference, however such a small change is unlikely to be of any clinical significance to a group. The term “personal significance”<sup>56</sup> takes account of the extent to which—regardless of numbers—a treatment remedies the life problems and consequent presenting clinical complaints that it causes.

In clinical psychology the “reliable change index” is used commonly to define what is considered to be 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 that procedure for percentage syllables stuttered data before and after treatment, and this method has been demonstrated.<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 that paper<sup>60</sup> argue—contentiously—that this can be done for percentage syllables stuttered scores by using 0.5 %SS as the cut-off score for normal 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 taken into account when evaluating the results of the trial.

One approach to dealing with clinical trial drop-outs is “intention to treat” analysis.<sup>61</sup> This means analysing trial outcomes of participants according to the treatment group they were randomised, regardless of whether they completed treatment or what treatment they completed.<sup>†</sup> One way to do this is “last observation carried forward.” This means that if a participant drops out, for example after collection of pre-randomisation speech measures, then those pre-randomisation speech measures are included as post-randomisation measures. This provides for 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 are 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 effective 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.

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<sup>†</sup> Sometimes participants “drop-in,” which means they receive the experimental treatment even though they were not randomised to receive it.

## FINDING STUTTERING RESEARCH TO INFORM EVIDENCE-BASED PRACTICE

### Finding clinical trials as they are published

Thousands of research papers have been published in scientific journals about stuttering, and hundreds are added each few years. Clearly, it is challenging for clinicians to keep up with such a burgeoning body of literature. However, based on the previous argument about the fundamental importance of clinical trials to everyday clinical practice with stuttering, it seems reasonable that finding and reading clinical trials from within that emerging literature should be a priority. There are several databases of scientific research that can assist clinicians to do that.

#### *Step One: Set up regular database email alerts*

Clinicians who have access through their workplace to a library with research databases can arrange for those databases to send regular email alerts about publications dealing with stuttering (and, of course, any other professionally pertinent research topics). For clinicians without institutional access to databases, there are freely available databases that can send regular email alerts.<sup>62,63</sup>

The most useful search string is “stutter\* or stammer\*” in the title and abstract fields.<sup>†</sup> Adding the term “trial” or “clinical trial” to the search string will not be useful, because clinical trials may not use the term “clinical trial” in either the title or the abstract.

It is arguably best to have email alerts sent once per week, for two reasons. First, it spreads out the workload sorting through publications to find which are clinical trials. Second, if a convincing clinical trial is published, then it is best to know about it immediately because it could influence clinical practices.

#### *Step Two: Scan publication titles*

When an email notification indicates there has been a publication dealing with stuttering, the titles can be scanned for any that obviously are not clinical trials. For example, titles such as “eye gaze patterns during social interactions with stuttered speech,” and “interhemispheric signal processing with stuttering” can be excluded right away during a search for clinical trials.

#### *Step Three: Read abstracts of possible trials*

If a title that comes up in a search that looks as if it might be a clinical trial, the abstract of the report can be inspected to see if it may have been a clinical trial. Examples of titles that suggest clinical trials are “Intensive stuttering modification therapy: multidimensional assessment of treatment outcomes”<sup>64</sup> and “Evaluation of a stuttering treatment based on reduction of short phonation intervals.”<sup>45</sup> The latter report contained the following,

All speakers achieved stutter-free and natural-sounding speech during within- and beyond-clinic speaking tasks at the completion of Maintenance. All were tested 12 months after completion of Maintenance. (p. 1229)

This strongly suggests that the report might qualify as a clinical trial that needs to be read carefully. On the other hand, the abstract of another paper<sup>41</sup> contains

The study involved assessment of the children's speech fluency and a client satisfaction questionnaire that sought parents' opinions about which aspects of the treatment were beneficial. ... Responses to the questionnaire indicated that treatment helped families learn about stuttering and about strategies that facilitate children's fluency. Evaluation of the children's fluency revealed that all participants achieved improved fluency at the conclusion of treatment and at long-term follow-up. (p. 118)

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<sup>†</sup> If a database will not accept the “\*” truncation character, “stutter,” “stuttering,” “stammer,” and “stammering,” will need to be entered separately.



This gives no indication of whether, according to the previous clinical trial definition, this could be considered a clinical trial. However, it might well be, and it needs to be read in detail to make a final determination. As it turns out, this is not a clinical trial according to that definition, because it was based on retrospective file audit data rather than prospective, beyond-clinic speech measures.

*Step Four: Read the clinical trials*

When it is clear that the report is a clinical trial of a stuttering treatment, the paper can be read in detail. During that reading clinicians can form a view of how, if at all, the trial could influence clinical practices. Part of that view will be the phase of clinical trial development. To reiterate, a Phase I nonrandomised clinical trial with few participants will be less compelling than a Phase III randomised trial with many participants.

The burden of work in all this so far is reasonable. Based on current publication rates, it is extremely unlikely that in the near future more than 10 clinical trials would be published in any one year. If 1 hour is devoted to reading each clinical trial, the burden of work over an entire year keeping up with stuttering clinical trials would be 10 hours at most.

*Step Five: Read as many other stuttering treatment reports as possible*

Steps One to Three will identify abstracts of stuttering treatment reports that are not clinical trials but are basic research or treatment outcome research that can usefully inform evidence-based treatment practices. This increases the workload for a clinician who treats stuttering. However, it has been argued that:<sup>32</sup>

even the most complex stuttering treatment journal article can be assessed ... in less than approximately an hour. Multiplied across many articles, this is an important time commitment, but it is not unreasonable, especially considering the alternative of continuing to spend time providing ineffective or otherwise less than ideal treatment. (p. 134)

## 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 treatments 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 EARLY STUTTERING TREATMENTS

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#### CLINICAL FEATURES OF EARLY STUTTERING

##### *Parent access to children*

Formal education in most English speaking countries begins at age 5 or 6 years. Clinically, the key feature of this age is that, compared to the formal schooling period of life, parents have more access to their children. It is true that many children for some part of their early life will attend pre-school, kindergartens or day care centres for all or part of the week. Generally speaking, however, one or both parents have more access to children than they do during later stages of formal education. Parent access to children during each day is a clinically central feature of early stuttering intervention. Parents do the three treatments outlined during this lecture when they are with their children during each day.

##### *An age of stuttering tractability*

Early stuttering appears to be extremely tractable. So tractable, in fact, that children may recover naturally without any formal treatment, as discussed during Lecture Two. It is possible that early treatment facilitates natural recovery.<sup>1</sup> As discussed during Lecture Nine, it seems that stuttering starts losing clinical tractability during the school-age period of 6–11 years. A review of treatment reports available at the end of the last century<sup>2</sup> concluded about the need for early intervention that “treatment after more than 15 months have elapsed does not appear to have been as effective ... as treatment initiated sooner” (p. 223).

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

Lecture Two overviewed the educational and occupational limitations often encountered by those who stutter, and their likely link to social anxiety. As discussed during Lecture Ten, there is good reason to believe that negative peer conditioning during the pre-school years is implicated in the origins of lifelong social anxiety with stuttering. Considering that, and considering that the disorder is at its most tractable during the pre-school years, treatment during that period of life is clearly the best option.

#### EARLY INTERVENTION WITH TELEPRACTICE

##### Telepractice

Current early stuttering interventions were developed for the traditional format of weekly clinic visits. The term telepractice, or telehealth, telemedicine and telerehabilitation that are sometimes used, refers to technology use to treat clients when they are not in the clinic. A review paper has overviewed telepractice in speech-language pathology with specific reference to stuttering treatment.<sup>3</sup> Professional speech-language pathology associations have overviewed telepractice service provision and associated professional issues with it.<sup>4,5,6</sup>

Telepractice is currently accessible with laptops and webcams, and with Internet transmission rates constantly improving, and with smartphone Internet accessibility. The number of households with Internet connections is increasing rapidly.



## Advantages of telepractice early stuttering intervention

### *Limited infrastructure needed*

The traditional clinical infrastructure is not needed for telepractice. All that is needed is a workspace, an Internet connection, and a laptop with a webcam. Webcam software is downloadable free of charge for clients and clinicians. And 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 pre-school child for travel to the clinic, and either arrange childcare for siblings or bring them along. This removes much of the effort parents need to expend to do the treatment with their children. There is evidence that parents can find this to be a significant burden with doing the treatment.<sup>7</sup> A significant clinical advantage of telepractice is that the clinician sees the parents doing treatment with their children in their own homes. Not only from the perspective of common sense, but also in terms of generalisation theory,<sup>8</sup> that makes clinical sense.

### *Families isolated from in-clinic services*

Telepractice is useful for the many families who are isolated from in-clinic treatment services. In large and sparsely populated countries, there will be many families who are isolated from such orthodox services. In Australia, for example, one third of families live rurally, and, apart from geographical isolation, such isolation presents many barriers to treatment access.<sup>9</sup>

### *Facilitates specialisation*

Telepractice facilitates clinical specialisation. It enables a specialist clinician to treat children who are located anywhere in the country. Additionally, children can be treated in any other country in a similar time zone to where the clinician is located. With flexible working hours, a clinician can treat children who are located in a country with a different time zone. So telepractice can facilitate a clinician becoming known nationally and internationally as a specialist with stuttering.

## THREE EARLY STUTTERING TREATMENTS SUPPORTED BY CLINICAL TRIALS

Three early stuttering interventions that have clinical trial evidence to support them using the definition of a clinical trial presented during Lecture Five—either Phase I, Phase II or Phase III evidence—are now overviewed. Those treatments are the Lidcombe Program, the Westmead Program, and two conceptually similar treatments: 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>10</sup> During the next lecture the clinical research that supports the three treatments is 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 during 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>11</sup> which adapted a technique developed a decade earlier.<sup>12</sup> The experimenters set up an illuminated puppet that conversed with children who stuttered. During the experimental conditions the light was turned off during moments of stuttering, effectively making the puppet disappear. The experimenters showed that this ingenious application of verbal response contingent stimulation successfully controlled the

stuttering of two pre-school children. That control generalised beyond the laboratory and was maintained for around 1 year.

#### *Resource materials*

The Lidcombe Program Treatment Guide<sup>13</sup> is a downloadable clinical reference for the treatment which can be found at the website of the Australian Stuttering Research Centre. At that website<sup>14</sup> there is a downloadable clinical severity rating (SR) chart for parents and clinicians. It is reproduced at Appendix One to this lecture. The website also contains a downloadable pamphlet about the treatment for parents, in 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>1,15</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>16</sup> and another is presented with case studies of its use.<sup>17</sup>

The Lidcombe Program Trainers Consortium<sup>18,19</sup> has members in 12 countries and provides postgraduate training for the treatment. That training involves two days of instruction and demonstration, often with subsequent clinical follow-up. It is recommended that clinicians do not attempt the Lidcombe Program without Consortium training, particularly in light of Phase IV evidence, discussed during the next lecture, that such training contributes to favourable community outcomes with the treatment.<sup>20</sup> The Lidcombe Program is endorsed by the professional associations of several countries.<sup>21,22,23</sup>

## Overview

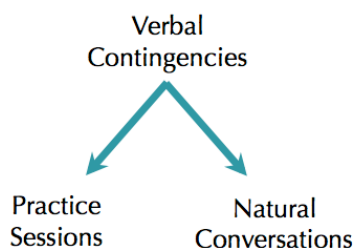
### *A behavioural treatment*

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 the training and supervision of a clinician. It involves parents giving their children verbal response contingent stimulation—verbal contingencies—for not stuttering and for stuttering. They do that during practice sessions with their children designed specifically for that purpose, and during naturally occurring conversations with their children. On most occasions it is parents who give the treatment to their children, but sometimes it is caregivers.



### *Clinical measurement*

Regular measurement of children's stuttering severity occurs during the Lidcombe Program with the SR scale described during Lecture Four: 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 9 = *extremely severe stuttering*. Parents and clinicians use the SR scale during treatment.



*Parents visit the clinic each week*

During each weekly visit the clinician teaches parents how to do the treatment and ensures that it is being done properly. The treatment guide<sup>13</sup> specifies what occurs during each clinic visit, 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 visit: 0 = *no stuttering*, 1 = *extremely mild stuttering*, 9 = *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 weeks:<sup>13</sup>

- (1) parent SRs of 0–1 during the week preceding the clinic visit with at least four of those seven SRs being 0, (2) clinician SRs of 0–1 during the clinic visit. (p. 10)

*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:<sup>13</sup>

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)

*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, in reality, attained the SR treatment goals just outlined. For example, parents might give an average SR of 0.3 for the week before a clinic visit 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 such problems. To quote from the treatment guide:<sup>13</sup>

During the first clinic visit, after the clinician has explained the SR scale, the parent or the clinician, or both, converse with the child for a few minutes 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 visits begin with the parent conversing

<sup>†</sup> Prior to 2015 the Lidcombe Program used a 1–10 SR scale, and publications before then contain that version of it.

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)

#### *Web based severity ratings*

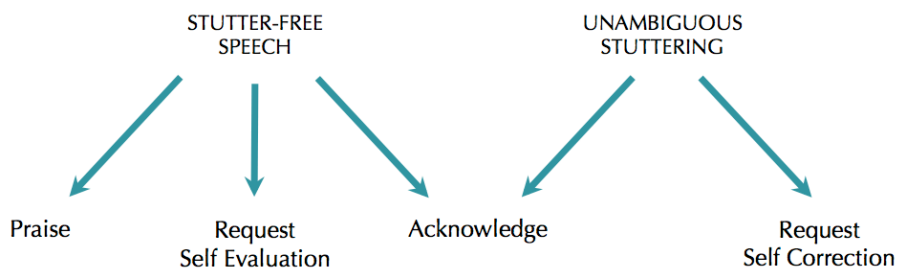
It is not necessary for parents to bring in hand-graphed SRs each week on a hard copy. A disadvantage of that procedure is that clinicians cannot monitor for whether parents are following instructions properly and recording a score at the end of each day. Sometimes, parents are not compliant with that instruction and will wait a few days before recording scores. This problem can be obviated, along with the need for hard copy, by using an Internet platform for SR scores that dates each entry<sup>24</sup> (see Lecture Four).

#### **Percentage syllables stuttered (%SS)**

There has been a recent recommendation, with theoretical and empirical justification, for why %SS is no longer recommended as part of the Lidcombe Program.<sup>25</sup> However, for reasons outlined in that publication, some clinicians 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 those are verbal contingencies for stutter-free speech, and two are for unambiguous stuttering moments. The essential Lidcombe Program verbal contingencies are overviewed in the figure.



#### *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 pre-school children.

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 essential also that parents are genuine with their praise and also 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 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 is “no.” Or, a parent could say “did you say all that smoothly?” and the expected response is “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 interrupted 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 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 "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 or on most stuttering moments, unless the child has only a few of them each day, which occurs toward 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 pre-school children receiving the Lidcombe Program, in particular, will sometimes spontaneously self evaluate their speech as stutter free, saying something like "I didn't do any bumps." In which case a parent may say 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 is 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 parent 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 visit. Naturally, this is a desirable thing to be happening and is 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 that is, at each clinic visit, 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 visits. Otherwise, the treatment process will not work properly if parents continue to do verbal contingencies incorrectly.

#### *They must be a positive child experience*

Nor will the Lidcombe Program treatment process work properly if verbal contingencies amount to a negative experience for the child. They 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 they can be sure that the child is receiving supportive and enjoyable verbal contingencies. Otherwise, during clinic visits it will be obvious that the child is not happy with the treatment, with predictable clinical results.

### **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, 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 length*

Lecture One presented research evidence that stuttering increases with increasing syntactic complexity and utterance length, and that those findings have been replicated with children. Clinicians can use that information to teach parents to manipulate those variables when giving verbal contingencies during practice sessions to minimise the occurrence of stuttered utterances if needed. With such manipulation, treatment during practice sessions can involve eliciting a range of utterance lengths and 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 length 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: meal times, in the bath, on the way to pre-school, in the park with the family, shopping, and so on. Eventually, verbal contingencies during natural conversations replace verbal contingencies during practice sessions, and the latter do not occur at all.

*Their purpose*

The fundamental clinical premise of the Lidcombe Program, based on laboratory research, is that parent verbal contingencies are the active treatment agent. So, when the clinician feels it to be appropriate, it is logical for parent verbal contingencies to occur during natural conversations with children.

**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 pre-schoolers, it does occur after the Lidcombe Program. In fact, half the children in one report<sup>26</sup> showed some transient signs of stuttering a mean of 5 years after their treatment began. So, the third purpose of Stage 2 is, after having detected any such signs, to prevent relapse from occurring.

*Treatment goals for Stage 2*

To progress to Stage 2 children need to meet the criteria mentioned earlier for three consecutive weeks:<sup>13</sup>

- (1) parent SRs of 0–1 during the week preceding the clinic visit with at least four of those seven SRs being 0, (2) clinician SRs of 0–1 during the clinic visit. (p. 10)

*Performance contingent maintenance*

The idea of a performance contingent maintenance schedule was introduced to stuttering treatment, and its potential benefits were shown in 1980.<sup>27</sup> It amounts to the parent and child returning to the clinic and having to sustain treatment targets for increasingly longer intervals; two visits 2 weeks apart, then two visits 4 weeks apart, followed by the same thing at 8 and 16 weeks between visits. If the child does not meet the Lidcombe Program treatment criteria at any visit, the parent and child return to the start of the sequence. Stage 2 normally takes a year or more. The importance of doing this

procedure is shown by a report that half of children during Stage 2 fail to meet treatment criteria at least once during Stage 2.<sup>28</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 serious risk that relapse will occur. The researchers who published a long-term clinical follow-up<sup>26</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.

#### **The 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>29</sup> Appendix Two shows the most common problems that needed to be solved.

## **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>13</sup> That guide specifies what occurs during each clinic visit, 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. Those two features of the treatment—adapting it for each family and being sure that parents are doing it correctly—are essential for it to succeed.

### **Limitations**

#### *It cannot be used 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 discussed during the next lecture, it is common for clinicians to delay treatment for a period after onset. As noted during Lecture Two, many children begin stuttering prior to 30 months of age. In the event that 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, during 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*

There is a safety issue with the Lidcombe Program connected with its use of parent verbal contingencies. Research has revealed the possibility, as mentioned earlier, that children could react negatively to verbal contingencies.<sup>107</sup> It is possible therefore, without proper clinician management to prevent such an event, for a parent to 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 multifactorial models described during Lecture Three. To reiterate briefly, this perspective states that what triggers stuttering and sustains it subsequently is found in predisposing motor, physiological, language and developmental child variables and the way they interact with their living environments. None of those 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 that 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<sup>‡</sup> overviewing the multifactorial model on which this treatment is based, that was presented during Lecture Three

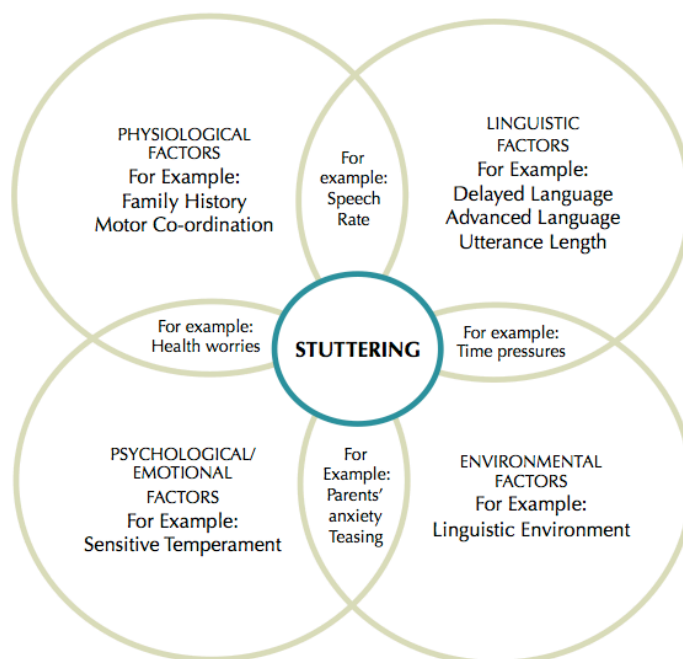
#### *Other treatment influences*

One of the developers of the treatment has noted that<sup>30</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)

#### *Diametrically opposite approaches*

Palin Parent-Child Interaction Therapy includes advice to parents<sup>31</sup> that "while your child will probably be more fluent if you ask them to say the problem word again, this is unlikely to help him/her the next time he or she tries to say the same word." This is more or less what the Lidcombe Program does, and so this highlights the different approaches of the two treatments. These differences



<sup>†</sup> Thanks to Alison Nicholas at the Michael Palin Centre, London, for assistance with preparing this description of the treatment.

<sup>‡</sup> Adapted and reproduced with permission: the Michael Palin Centre, © 2014.

were discussed with one of the Palin Parent- Child Interaction Therapy developers in a journal article.<sup>30</sup>

#### *Resource materials*

Two journal reports contain overviews of the treatment.<sup>32,33</sup> A slightly longer overview is in a book chapter,<sup>34</sup> and there is a comprehensive manual available.<sup>35</sup> Additionally, the Michael Palin Centre conducts a 3-day training for the procedure,<sup>36</sup> which is available only in the United Kingdom. A web page at the Michael Palin Centre website contains general advice to parents of stuttering pre-school children.<sup>31</sup>

#### *General advice to families of stuttering pre-schoolers*

The advice to parents of children who stutter at that location<sup>31</sup> includes the following:

- (1) Find periods during each day to give children complete attention in a relaxed atmosphere
- (2) Focus on what children say, not the way they say it, and do not look away when they are stuttering
- (3) Reduce parent speech rate to create a relaxed atmosphere
- (4) Reduce the number of questions asked of children and provide time for answers
- (5) Allow children to finish utterances rather than finishing them
- (6) Explain to the family the importance of conversational turn taking
- (7) Praise things about children that are independent of speech
- (8) Discipline a stuttering child identically to any other child
- (9) Introduce routine and structure into daily life
- (10) Ensure sufficient sleep and an adequate diet

## **Overview**

#### *Pre-treatment assessment*

The treatment begins with a detailed assessment that takes account of general speech and language development in addition to stuttering. The assessment is designed also 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. Additionally, 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>30</sup>

And, stated at another source:<sup>34</sup>

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)

And at another source:<sup>33</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: psychological, physiological, language, living environment. The clinician has available 40 “interaction strategies” classified within 12 categories, outlined in Chapter Six of the treatment manual:<sup>35</sup>

- (1) Following the child’s lead in play
- (2) Letting the child solve problems for himself
- (3) Using more comments than questions during conversation
- (4) Complexity of questions at child’s level
- (5) Using language which is appropriate for the child’s level
- (6) Using language which is semantically contingent on the child’s focus of attention
- (7) Using repetition, expansion and rephrasing of the child’s utterance
- (8) Giving the child time to initiate, respond and finish his talking
- (9) Matching the parent’s rate to the child’s rate
- (10) Using pausing before and between utterances
- (11) Use of eye contact, position, touch, humour and/or surprise
- (12) Using praise and reinforcement (p. 91–125)

Additionally, there are 19 “family strategies” outlined in Chapter Seven of the treatment manual:<sup>35</sup>

- (1) Managing two languages
- (2) Openness about stammering
- (3) Building confidence
- (4) Giving children feedback
- (5) Sincerity
- (6) Consistency
- (7) The language of praise
- (8) Reactions to praise
- (9) Helping parents to build up their child’s confidence
- (10) Turn-taking
- (11) Dealing with feelings
- (12) Difficulties with separation
- (13) High standards
- (14) Helping parents to manage their child who has very high standards
- (15) Sleep
- (16) Behaviour management
- (17) Routines
- (18) Pace of life
- (19) Emerging issues (p. 127–168)

The treatment manual<sup>35</sup> outlines “child strategies” in Chapter Eight, stating that “our research has shown that most children achieve fluency with the interaction and family strategies ... However some children’s fluency continues to be a cause for concern and we introduce direct fluency therapy at this stage” (p. 169). These strategies incorporate speech restructuring treatment components:

- (1) Rate reduction
- (2) Pausing to think
- (3) Easy onset
- (4) Being more concise

(5) Eye contact/focus of attention (p. 169)

## The treatment process

### *“Special time”*

The treatment incorporates “special time,” which is a 5-minute period that each parent spends individually with the child three to five times per week. The purpose of special time is to provide a comfortable environment in which parents can practice the targeted interaction changes. It is expected that the changes to parent interactive style will generalise to beyond these talking times, but nonetheless they continue to occur throughout the treatment.

### *Clinic visits*

The treatment involves six weekly, 1-hour clinic visits. The format of each clinic visit is consistent, with the exception that during the first clinic visit the results of the assessment are conveyed to parents and the routine for special time is established for the family. During special time parents keep a diary about the activity conducted and the targets that they implement, and this diary is presented to the clinician for discussion at the start of each session.

Both parents are required to attend each clinic session, during which a version of special time is conducted in the clinic and is video recorded. The clinician is nondirective during the treatment, and parents are encouraged to select their own treatment targets based on their observations of the within-clinic video. Parents are encouraged to identify the interaction styles that they are already using to support the child’s fluency and they select an interaction style that they would like to perform more often, and the introduction of any new targets is discussed with the clinician.

### *The consolidation period*

After the six weekly clinic visits, there is 6 weeks of a “consolidation” period, which occurs entirely at home. The purpose of this is for parents to consolidate the skills they have learned and generalise them to the home environment. There is no mention in any of the documentation about the treatment stating that targeted family interaction changes should generalise to beyond the home environment. However, the clinician may involve nursery or school staff as appropriate.

During the consolidation period parents send their special time diaries to the clinician each week, and they receive written feedback from the clinician. An example of such a diary is presented in the treatment manual.<sup>35</sup> Subsequent to the 6-week consolidation period, review clinic visits are scheduled at 3 weeks, 3 months, 6 months and 1 year.

### *Standard treatment period*

Unlike the Lidcombe Program, Palin Parent-Child Interaction Therapy has a specified number of six clinic visits. It does not involve speech criteria that are used to establish an end to the treatment, although %SS measures are collected at assessment, pre-treatment and post-treatment, along with parent rating scales. The clinician considers these measures when making a decision about the end of treatment.

### *Treatment flexibility*

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>32</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:<sup>35</sup>

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. 84)

## TREATMENTS BASED ON MULTIFACTORIAL MODELS:

### II. RESTART-DCM<sup>†</sup> TREATMENT

#### Background

This treatment has much in common conceptually and procedurally with Palin Parent-Child Interaction Therapy. It also has popularity in common with that treatment, being widespread in the Netherlands since the 1980s and “taught to Dutch students of speech therapy for the past 25 years” (p. 2).<sup>37</sup>

#### Overview

In common with Palin Parent-Child Interaction Therapy, the treatment is a hybrid involving strategies derived directly from multifactorial models of stuttering causality and speech restructuring techniques. As stated in the treatment manual<sup>37</sup>

The RESTART-DCM approach is never limited to simply providing advice to the parents. Depending on what is found to be necessary, the speech (stuttering) therapy provided will focus on behaviour changes, coping with emotions and skills training. If lowering the demands and promoting the capacities should fail to resolve the stuttering problem to a satisfactory extent, speech fluency may be worked on directly by *modelling* slower, more relaxed, smoother speech (p. 4)

Additionally, all children are given an oral motor assessment.<sup>38</sup> If that assessment “should reveal that the oral motor skills are insufficient, reinforcement of the motor skills is a relevant therapy goal” (p. 12) and the child is treated with a method involving speech motor drills<sup>39</sup> in addition to the basic procedures.

#### Assessment

The treatment manual<sup>37</sup> outlines five standard tests of language, articulation and oral motor function. Additionally, two 10–15 minute video recordings are made of the child and parents playing together in their customary fashion. This interaction is then scored using a form in Appendix One of the treatment manual, using the following categories. The italics below are any “unfavourable behaviour” (p. 19) that is noted on the video:

- (1) Questions parent to child: *many; open; a commanding tone, or with little time for the child to answer*
- (2) Turn-taking behaviour: *talking simultaneously, interrupting; interaction times are too short*
- (3) Parent response to stuttering: *negative verbal reaction to the stuttering; negative non-verbal reaction to the stuttering*
- (4) Parent(s) linguistic behaviour: *introduce a new topic; correct child’s verbal behaviour; make utterances that increase time pressure*
- (5) Articulation and/or speech rate
- (6) Other parental behaviour: *give negative attention; show directive action* (p. 19)

In relation to (2) and (5) it appears that the treatment is not, strictly speaking, a clinical application of a multifactorial model of stuttering because the treatment is not different for every child: all parents speak with reduced speech rate to their children and with increased interturn speaker latencies.<sup>37</sup>

A rule of thumb to establish the right speed is to speak just as slowly as the child does when speaking fluently, unless the child has a relatively rapid rate of speech, i.e., > 3.5 syll /sec. In that case, the parent must learn to speak at slow-to-normal speed ... (p. 8)

<sup>†</sup> Rotterdam Evaluation Study of Stuttering Therapy-Demands and Capacities Model

Parents will rarely speak with a lower articulation rate than their children, so they routinely need to reduce their rate during the treatment.<sup>40</sup> The same applies to parent interturn speaker latencies,<sup>40</sup> because parents need to have latencies of 1–2 seconds during conversation with their children with “definitely no overlapping speech” (p. 8), which rarely occurs naturally.

### The treatment process

According to the treatment manual,<sup>37</sup> parents and children attend the clinic for an hour each week initially, but there is flexibility about the duration. Generally, after four visits parents are invited to attend a clinic session during which the child is not present.

During the first clinic visit the clinician explains causal factors for stuttering and discusses stuttering with parents, with that discussion supplemented by written material presented in Appendix Two of the treatment manual. During the first session parents are instructed to provide “parent-child special times” for 15 minutes per day at least 5 days a week. As treatment progresses those times are used to practise skills to lower demands and reinforce capacities according to clinician guidance. Parents keep a log of these special times.

Typical treatment sessions involve the clinician observing the parent playing and talking with the child, discussing progress during the previous week, and having the parent demonstrate treatment procedures that were used during the previous week. The clinician then outlines changes to clinical procedures for use during the coming week, demonstrates them to the parent, and has the parent attempt the procedures.

The following components of the treatment are outlined in the manual:<sup>37</sup>

- (1) Reducing motoric demands
- (2) Reducing linguistic demands
- (3) Reducing emotional demands
- (4) Reducing cognitive demands
- (5) Reinforcement of the speech motor capacity
- (6) Reinforcement of linguistic capacity
- (7) Reinforcement of the emotional capacity
- (8) Reinforcement of the cognitive capacities
- (9) Direct therapy with children aimed at more fluent speech (p. 7–16)

When the following program criteria are met, a 24-month maintenance phase begins, comprising three 30-minute clinic visits each month followed by one visit every 3 months for 21 months. The program criteria are<sup>37</sup>

The child has normal-fluent speech (very young children for approximately six weeks and older children [aged 4 1/2 -6] approximately 3-4 months) or exhibits only incidental disfluencies that are minimally abnormal (occasional repetitions with usually one iteration).

The parents implement a fluency enhancing environment or the speech therapist/fluency expert judges that the parents can maintain the rest of the modification on their own.

The child’s speech is acceptable to the parents, the child and the speech therapist/fluency expert.

The parents know what to do if a relapse should occur. (p. 16)

## CLINICAL STRENGTHS AND LIMITATIONS OF TREATMENTS BASED ON MULTIFACTORIAL MODELS

### Strengths

*They can be used for immediate early intervention*

With these treatments, the child does not have to do anything at all. It is only the parents who do the therapy. It is a completely passive treatment from the perspective of children. As such, these treatments are often described as indirect. Because of this they are suitable for children who stutter at any age.

### Limitations

*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 that more than 60 therapy strategies are involved with each of the treatments. Data about the matter are limited, but one of the clinical trials of Palin Parent-Child Interaction therapy<sup>32</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. And as discussed earlier, it seems that there are consistent elements in RESTART-DCM treatment that are used for every child who is treated: reduced parent speech rate and interturn speaker latency.

*Issues with the underpinning theoretical model*

A treatment based on a theoretical model of the nature of stuttering will be questionable 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 both involve the clinical option of a variant of speech restructuring if needed. Also, the latter treatment involves a speech motor training program in the event that 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>41,42</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 pre-schoolers*

During the early 1980s some researchers looked for clinically useful effects when pre-school children spoke in rhythm.<sup>43</sup> The children in that 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 a sequence from three single-syllable phrases, to four-six 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>44</sup> showed with 9–11 year old boys that instructions were not necessary for them to decrease stuttering in the presence of a metronome. The experimenter 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 most suggestive of clinical value for syllable-timed speech with children.

#### *Resource materials*

At present, the only Westmead Program resource materials are the published Phase I and Phase II clinical trials overviewed during the next lecture.<sup>45,46,47</sup>

### **The treatment process**

#### *Overview*

The Westmead Program directs parents to encourage children to use syllable-timed speech—“robot talking”—during everyday conversations. The aim is to achieve normal 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 practice syllable-timed speech. Parents occasionally praise their child for using this speech pattern. Parents prompt their children to use syllable-timed speech occasionally 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 visit the clinic each week*

As with all evidence-based early stuttering treatments, parents and children visit the clinic each week. During each weekly visit 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 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.

Treatment criteria are specified with two measures. The first of these is %SS measured by the clinician at the start of each session during conversation with the child or while the child converses with a parent. Stage 1 concludes, and Stage 2 begins, when these measures are below 1.0 %SS in the clinic for three consecutive weeks.

The second measure for treatment criteria is the SR scale used with the Lidcombe Program. Stage 1 concludes, and Stage 2 begins when, for three consecutive weeks, parent SRs are 0–1 during the week preceding the clinic visit, with at least four of those seven SRs being 0.

#### *Stage 1*

Stage 1 of the treatment has two components. 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 parents, where necessary, to modify utterance length and grammatical complexity to make syllable-timed speech easier to learn. Generally, children learn to do the speech pattern quickly and are able to do it during conversation during the first few sessions. At this time 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 visits begin During Stage 1B.

#### *Stage 2*

When children attain the treatment criteria, Stage 2 begins, and the family makes visits to the clinic less frequently during a period of 1 year. During Stage 2 parents are instructed to gradually stop doing the practice sessions each day. In the event that during a Stage 2 clinic visit 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, this 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 visits 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 it. 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 wearing for them to sustain for long periods in order to obtain durable stuttering control.

## **SUMMARY**

The pre-school years are a time when stuttering is at its most tractable and when parents have optimal access to their children during daily life. Therefore early stuttering intervention is a desirable clinical option, either within-clinic or telepractice treatment. There are three treatment types for pre-school children 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 process and each has distinctive strengths and limitations.

# APPENDIX ONE

## LIDCOMBE PROGRAM SEVERITY RATING CHART

**Severity Ratings**

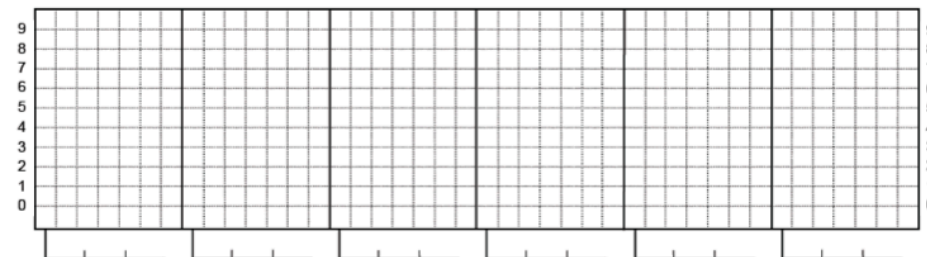
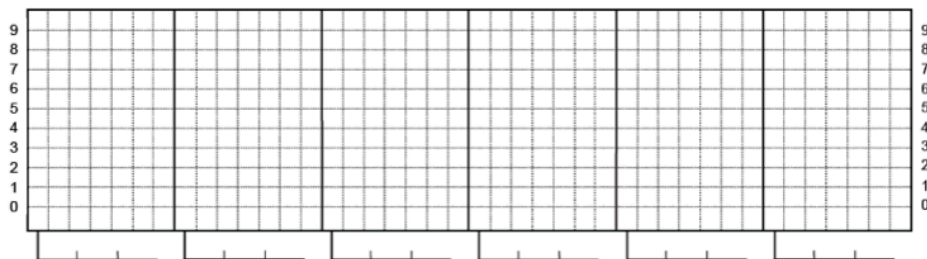
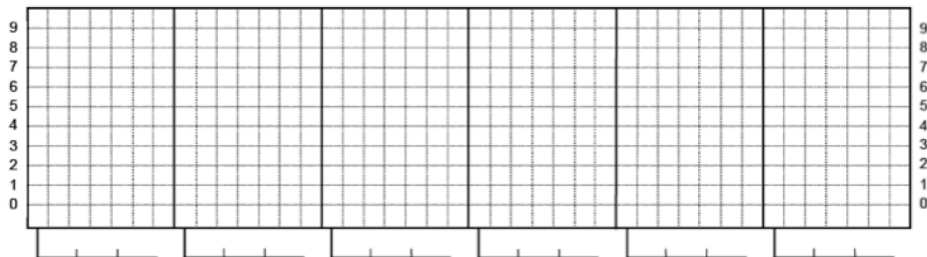
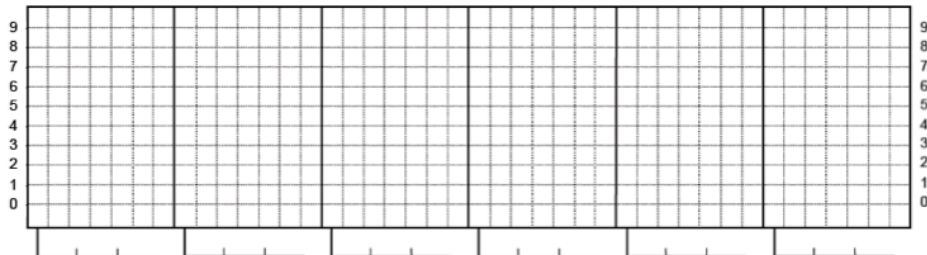
SR 0 = no stuttering

SR 1 = extremely mild stuttering

SR 9 = extremely severe stuttering

Name

Page





## APPENDIX TWO

### COMMON LIDCOMBE PROGRAM PROBLEMS<sup>29</sup>

<i>Speech measures</i>	<i>SRs not collected or collected infrequently. Parents unreliable using SRs.</i>	14% <sup>†</sup>
<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 during treatment.</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 many treatment goals for different disorders.</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 visits. 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%

<sup>†</sup> Percentage of consultations for which the problem occurred

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## LECTURE SEVEN

### THE EARLY 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 that format, the first Phase I non-randomised clinical trial with Australian children was published in 1990.<sup>1</sup> Subsequently, there was 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 years follow-up 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 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 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. That result was statistically and clinically significant.

A Phase III randomised controlled trial<sup>9</sup> compared a standard treatment arm with 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>



###### *Telepractice Phase I-II trials*

There have been three low-tech telepractice trials with Australian children using the telephone: two Phase I trials<sup>10,11</sup> and one randomised Phase II trial.<sup>12</sup> A Phase I trial of webcam telepractice has been published.<sup>13</sup> A Phase III randomised controlled webcam trial has been published, with an in-clinic and a webcam telepractice arm.<sup>14</sup> The design is known as a parallel, open plan, non-inferiority randomized controlled trial. Results showed no reason to believe that the webcam Lidcombe Program was less efficacious in terms of stuttering severity outcomes, or cost, than the clinic presentation. In fact, the webcam arm of the trial had 17% shorter treatment consultations 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. A clinical guide to conducting the Lidcombe Program by webcam is available.<sup>15</sup>

It is not clear at present where telepractice Lidcombe Program developments will lead. It could turn out that this treatment method will be suitable for the majority of families. On the other hand, that may not be so and the final place for telepractice Lidcombe Program intervention may be as part of a stepped care public health approach to early stuttering.

The stepped care model of healthcare delivery contains two fundamentals.<sup>16</sup> It provides the simplest and most cost efficient method of health care that is efficacious. 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 telepractice early stuttering intervention, they might then go to a clinic each week. Or an intervening step might be that telepractice Lidcombe Program 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>17</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 disorders,<sup>18,19,20,21,22</sup> but there seems to have been only one description of the stepped care concept applied to stuttering.<sup>23</sup>

#### *The Lidcombe Program in different cultures*

The treatment focuses 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 pre-schoolers,<sup>24</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 pre-school children were studied, one of whom was treated in Mandarin 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 overviewed 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 pre-school children.<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. 6)

#### *The Lidcombe Program with co-occurring speech sound disorder*

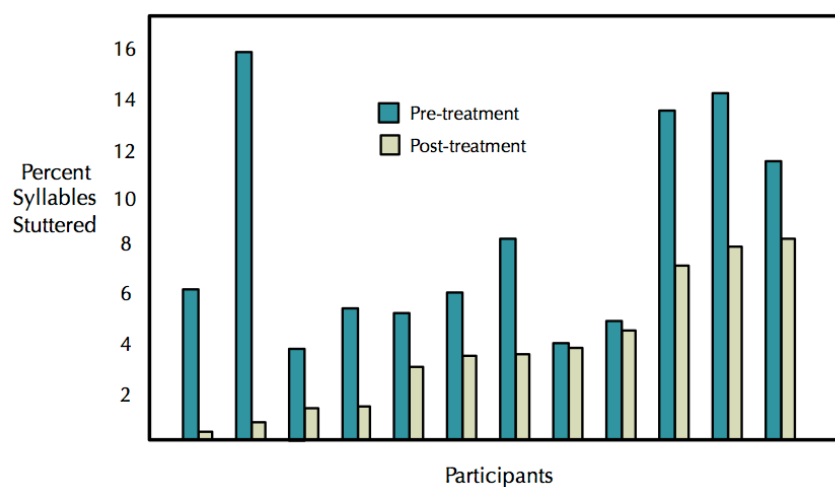
A Phase I trial<sup>25</sup> with five boys, ages 3–4 years, involved treatment for stuttering with the Lidcombe Program concurrently with treatment for speech sound disorder. The children were assessed at pre-treatment, at entry to Stage 2 of the Lidcombe Program, and 9 and 12 months after the start 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 Lidcombe Program treatment. Pre-treatment stuttering for the four children was in the range 2–15 %SS, and at 12 months post they were all below 1.0 %SS. At 12 months post, 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. 1).

**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>26,27</sup> The first trial<sup>26</sup> recruited nine families, of whom three dropped out, and the latter trial<sup>27</sup> recruited six children who were retained in the trial. The results across the two non-randomised trials are presented in the figure for the 12 children. For some of the children in the figure, follow-up data are for 6 months post-treatment,<sup>27</sup> and for some the follow-up data are 12 months<sup>26</sup> post-treatment.

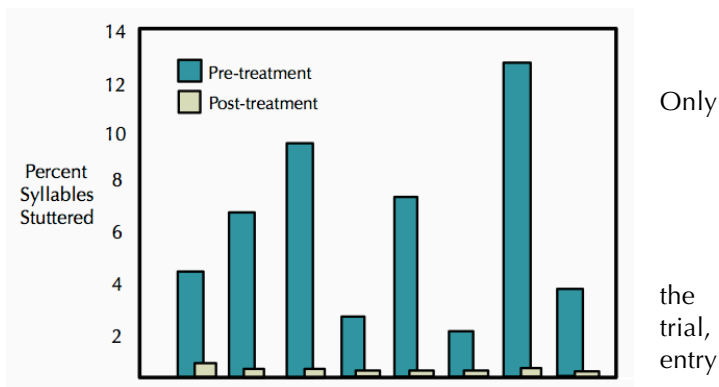


The first four children, on the left of the graph, achieved stuttering reductions to around 1.0 %SS or lower, but this was not the case for the other eight children. Two of them showed almost no stuttering reduction. Overall, the pre-treatment to post-treatment reductions of %SS were 64% for the 12 children. Considering that non-randomised designs overestimate effect sizes, this result arguably could be a reflection of natural recovery.

**The Westmead Program**

*In-clinic Phase I-II trials*

Subsequent to Phase I trials,<sup>28,29</sup> a Phase II trial<sup>30</sup> recruited 17 children. eight of the children completed the treatment. With that caveat in mind, along with the caveat about interpreting non-randomised evidence, the figure suggests that the treatment may have some merit. For eight children who completed the the mean post-treatment score at to Stage 2 was 0.2 %SS.



## CLINICAL TRIALS COMPARING TWO TREATMENTS

### Lidcombe Program compared to RESTART-DCM Treatment

#### *Method*

This is the largest randomised controlled trial reported for any stuttering treatment,<sup>31</sup> randomising 99 children to a Lidcombe Program arm and 100 children to a RESTART-DCM arm. 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>32</sup> and children in the latter group were treated with the RESTART-DCM manual mentioned during the previous lecture.<sup>33</sup> Children were followed up for 18 months after the start of treatment.

#### *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>34,35</sup> and “DCM based treatment training is included in the regular clinical education in the Netherlands” (p. 3).<sup>31</sup> Clinicians had a mean of 3.7 years experience with the Lidcombe Program and a mean of 15 years 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>36</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>31</sup> That 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 those recordings were during conversation 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>37</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, 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>38</sup> Additional secondary outcomes were a health-related quality of life measure (EQ-5D),<sup>39</sup> a measure of child attitude to communication (KiddyCat)<sup>40</sup> (see Lecture Ten), and three measures of child emotional and behavioural problems derived from the Child Behavior Checklist.<sup>41</sup>

#### *Primary outcome results*

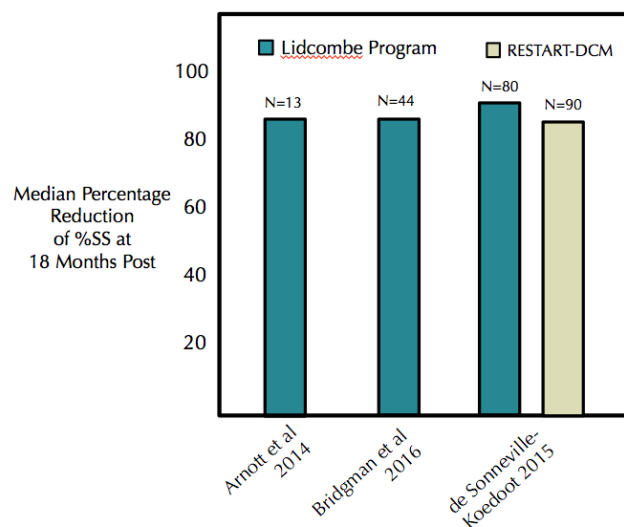
At 18 months after the start of treatment, 86 children remained in the Lidcombe Program arm and 91 remained in the RESTART-DCM arm. There were 76.5% of “non-stuttering” children in the Lidcombe Program group and 71.4% of “non-stuttering” children in the RESTART-DCM group. Those differences were not significant. Results remained nonsignificant when the cut-off %SS scores for “non-stuttering” were changed to 1.0 %SS and 2.0 %SS.

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. The reason is the width of the 95% confidence intervals for the mean values that are reported. There is a 95% chance that such confidence intervals contain the true mean. 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. 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. Arguably, the range of those confidence intervals includes differences that are clinically significant.

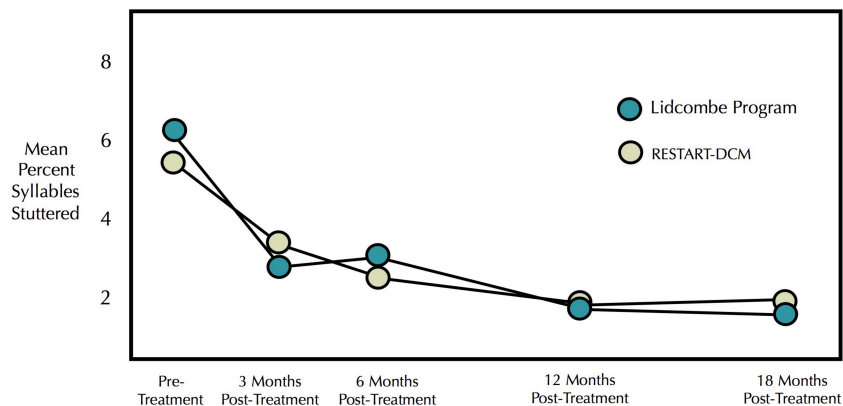
It would be useful to have some way of comparing the outcomes of the RESTART trial with the outcomes of the dedicated Lidcombe Program clinical trials discussed earlier. Any such comparison needs to be guarded, because the trials concerned were conducted in different countries, at different times, and 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. Arguably, a measure of percentage reduction of %SS scores from pre-treatment to 18 months post-treatment would go some way to offsetting any such reliability problems. So, the figure presents median percentage reductions for the RESTART trial and the data from the two standard treatment arms from Lidcombe Program randomised trials that presented data at 18 months post-randomisation.<sup>9,14</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 favor of the direct approach (LP), but the few significant interaction terms were deemed negligible due to their small effect sizes” (p. 11).<sup>31</sup> For the entire 18 months 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<sup>‡</sup> presents the %SS measures for the trial at pre-treatment, and 3, 6, 12, and 18 months after the start of treatment.

<sup>‡</sup> Adapted and reproduced with permission: De Sonnevill-Koedoot, C., Stolk, E., Rietveld, T., & Franken, M-C. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PLoS One*, 10, e0133758.



#### *An economic evaluation of the two treatments*

An economic evaluation of the RESTART clinical trial<sup>42</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>42</sup>

#### *A critique*

The RESTART trial attracted a negative critique<sup>43</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. 1). The treatment was criticised also because of the paradoxical criterion of  $\leq 1.5$  %SS as non-stuttering. The authors responded<sup>44</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.<sup>†</sup>

## TRANSLATIONAL RESEARCH

### The Lidcombe Program

One translational study,<sup>45</sup> an in-clinic Phase IV trial, 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 pre-school children. 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: pre-treatment stuttering severity, speech or language disorders in addition to stuttering, whether the clinicians had received training from the Lidcombe Program Trainers Consortium,<sup>46</sup> the duration of weekly clinic visits, and 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.

<sup>†</sup> These criticisms about the primary outcome seem 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.

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 are able to match those attained in clinical trials.

A study involving six community clinicians<sup>47</sup> evaluated the cost-effective rolling-group Lidcombe Program model used in a previous randomised trial.<sup>9</sup> Participants were 19 pre-school children, mean age 49.1 months, treated by six generalist clinicians in four Australian rural towns. Within clinic measures of percentage syllables stuttered were collected at pre-treatment and at 6 and 9 months after the start of treatment. Percentage syllables stuttered 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 (those benchmarks will be discussed shortly). The report was supplemented with a resource involving perspectives from the participating clinicians about the rolling-group treatment model.<sup>48</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 in order 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 that definition, with the exception that they are evaluations of parts of a treatment. Those reports might be termed randomised clinical experiments.

### The Lidcombe Program

#### *Sixteen weeks of Lidcombe Program compared to no treatment*

An experiment<sup>49</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 or 16 weeks of no treatment. One child dropped out of the treatment group. After 16 weeks of treatment, in everyday speaking situations, the Lidcombe Program children had 1.6 %SS and the control children 6.9 %SS. That 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>50</sup> randomised 29 pre-school children, four of them girls, to receive either 12 weeks of the Lidcombe Program 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, during everyday conversations, the children in the Lidcombe Program group had a mean 3.5 %SS and the children in the control group had a mean 5.8 %SS. That result was statistically and clinically significant.

#### *Twelve weeks of Lidcombe Program compared to RESTART-DCM*

As a preliminary to the randomised trial discussed earlier,<sup>31</sup> a Dutch study<sup>51</sup> randomised pre-school children to a Lidcombe Program group and a RESTART-DCM group. Thirty children were randomised, and 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. That result was clinically and statistically nonsignificant.

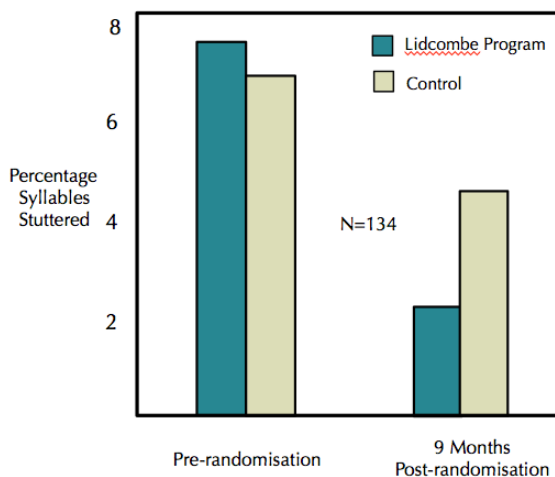
Interpreting their findings cautiously, and foreshadowing their later randomised trial, the authors concluded that “randomized controlled trials of LP versus DCM treatments are feasible” (p. 197).<sup>51</sup>

They also correctly pointed out that further study of the matter is necessary with control groups in order to obtain fully interpretable results.

#### *“Meta-analysis” of the Lidcombe Program*

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. However, the next best thing is a meta-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>7,12</sup> and two randomised clinical experiments.<sup>49,50</sup> The mean post-randomisation period for those reports is 6.3 months.

The results of this analysis<sup>52</sup> are shown in the graph. In total, it involved 134 children. 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<sup>†</sup> 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, meaning that there was an estimated 95% chance of the odds ratio being 2.7 or more.

It is necessary when interpreting this meta-analysis result to keep in mind 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”<sup>53</sup> (p. 251), which presumably means 21 months after completing Stage 1. Several other case studies have been reported for the Lidcombe Program with British,<sup>54</sup> Canadian,<sup>55</sup> and United States children.<sup>56</sup> 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. Two individual case studies of the Lidcombe Program have been

<sup>†</sup> See Lecture Five for more discussion of odds ratios.

reported with French Canadian pre-schoolers.<sup>57,58,59</sup> Those cases were consistent with a report of Malayasian children treated with the Lidcombe Program<sup>2</sup> that treatment in one language generalises to another language.

### A family-focused treatment approach based on a multifactorial model

There has been a case study report<sup>60</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.

Similar to the Palin Parent-Child Interaction Therapy treatment process, this family-focused treatment approach “typically consists of six to eight sessions, 45 min in length, scheduled once per week or every other week” (p. 120).<sup>60</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<sup>60</sup>

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)

Also consistent with Palin Parent-Child Interaction Therapy, and also RESTART-DCM Treatment, is that “direct fluency shaping and stuttering modification” (p. 119)<sup>60</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.<sup>60</sup>

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)

The specific “parent communication modifications” considered are<sup>60</sup>

(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)

The results of the report were fairly consistent with the two clinical trials of Palin Parent-Child Interaction Therapy,<sup>26,27</sup> indicating that six of 17 children “continued to stutter following completion of the parent-focused treatment” (p. 128).<sup>60</sup> Mean scores for “stuttered types of disfluencies per 100 words” (p. 126)<sup>60</sup> were 16.4 before and 3.2 after the intervention. Parents scored “how often the child was able to speak *without* [authors’ italics] stuttering” (p. 126–127)<sup>60</sup> 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.

## TREATMENT FIDELITY RESEARCH

### The Lidcombe Program

#### *Therapist drift*

Treatment fidelity refers to whether a treatment is administered as intended, and is an important consideration with treatment translation in general,<sup>61,62</sup> and the issue is thought to be important with stuttering treatment.<sup>63,64</sup> Departure from manualised procedures, or therapist drift<sup>65</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.

#### *Therapist drift in a translational study*

During the translational study outlined previously,<sup>45</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 treatment sessions and used 30-minute sessions instead. Also, fortnightly clinic visits occurred often instead of the prescribed weekly visits, sometimes because of clinician scheduling and sometimes because of client failures to attend for 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>66</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 that treatment fidelity problem affected outcome. Regardless, the authors concluded:<sup>45</sup>

The fact that only half the SLPs 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 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 in the present study largely chose not to adhere to this important treatment procedure. (p. 601–602)

#### *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 pre-school children during their treatment.<sup>67</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. That was the case also with verbal contingencies during natural conversations, although more contingencies for stuttering occurred in such situations.

The report had some sobering features, however. One parent continued with the treatment during practice sessions despite the child saying he did not like the activity. Another parent focussed 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 during treatment in practice sessions, such as praising stuttered speech.

To assist clinicians with ensuring Lidcombe Program treatment fidelity, the report<sup>67</sup> presents an empirically developed checklist of procedures for treatment during practice sessions, which focuses on the important issues of parent verbal contingencies. The paper contains two case histories of the checklist being used.

A larger study<sup>68</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 a survey study*

A survey of 277 Australian speech pathologists<sup>69</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 the Australian public health providers sometimes do 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, with one report of children waiting up to 12 months for treatment. Other problems identified included allocating the requisite time in school settings that provided treatment services. For the study sample, 23% of clinicians were located 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>70</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.

## **TREATMENT MECHANISMS**

As discussed during the previous lecture, treatments based on multifactorial models and the Westmead Program have transparent, putative underlying mechanisms for any treatment effects. Although there are no data at present to substantiate such notions, the former treatments might operate because multifactorial models are correct, and the Westmead Program might operate because of the acoustic effects of syllable-timed speech. However, there is no obvious mechanism that might explain how the treatment effects of the Lidcombe Program might occur.

It is reasonable speculation, regardless, that the treatment somehow rectifies problems with neural speech processing connected with stuttering (see Lecture Three). Perhaps an efficacious treatment such as the Lidcombe Program induces children to “adopt a compensatory neural growth pattern that successfully makes up for the deficient brain regions” (p. 77).<sup>71</sup> Another author has suggested a similar mechanism in terms of the malleable nature of the developing brain.<sup>72</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>73</sup>

This idea incorporates the well-known notion of “cortical plasticity.” Not only does the brain drive behaviour, but behaviour drives the brain. For example, learning to juggle can change gray matter structure in areas that support visual learning.<sup>74</sup> Another study showed changes in the occipito-temporal cortex after only seven days of juggling learning.<sup>75</sup>

### **The Lidcombe Program**

Several studies have attempted to find some explanatory underlying mechanism for the Lidcombe Program.

#### *Child and parent language*

One suggestion has been that children, or parents, might simplify their language production after the treatment. An initial report with nine parent-child pairs found that to not be the case,<sup>76</sup> 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>55</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>77</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>78</sup> was reported for 11 pre-school children prior to Lidcombe Program treatment, and 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>77</sup> perhaps children use a slightly different speech pattern post-treatment that controls stuttering. However, that report found no changes for vowel duration, intervocalic interval, voice onset time, or articulation rate.

#### *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 carries the assumption 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 experimenters 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. That result challenged the contribution of the verbal contingency *request self correction* to the efficacy of the treatment, and suggested the need for further research about the matter. This study supplements the study of parent treatment fidelity with verbal contingencies discussed earlier.<sup>68</sup> Together, the two studies raise an issue in need of resolution about how verbal contingencies contribute to Lidcombe Program treatment effects.

### **Treatments based on multifactorial models**

A review of the topic<sup>80</sup> and a subsequent report<sup>81</sup> found no reason to implicate unusual parent language behaviour with early stuttering, or any reason to believe that changing parent language behaviour may be clinically useful with early stuttering.

As noted in the previous Lecture, during RESTART-DCM treatment all parents 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 stuttering is of interest. A recent review<sup>82</sup> of five laboratory experiments of parent reduced speech rate with pre-school children<sup>83,84,85,86,87</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>84</sup> concluded that study of the effects of extended, everyday parent use of such techniques is warranted.



An experimental study of a 5 year 9 month old child who stuttered<sup>88</sup> involved increased interturn speaker latency for 15 sessions during a 7-week period at the family dinner table. The parents and his 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>89</sup> with three boys who stuttered involved three experimental sessions of “no interruption” for two of them during conversation with an experimenter. For one boy, age 6 years 2 months, a stuttering reduction of around 50% was observed, and no effect was observed for the other boy, age 5 years 6 months. A study of 27 pre-school children who stuttered,<sup>90</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%.

## TREATMENT SAFETY

### The Lidcombe Program

As noted during 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. That situation can become clinically problematic if the clinician does not recognise it and respond appropriately. Indeed, the early development of the Lidcombe Program prompted concerns that the treatment might send an overall negative message to children affecting their self esteem, and establishing unhelpful cognitions.<sup>91,92,93</sup>

In response to those concerns, the Lidcombe Program developers verified with eight pre-school children that the treatment is psychologically safe.<sup>94</sup> Measures with the Child Behaviour Checklist<sup>95</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 treatment. Additionally, the Attachment Q-Set, 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>31</sup> which reported some suggestion of post-treatment improvement for Child Behavior Checklist and KiddyCat scores after Lidcombe Program treatment.

### Treatments based on multifactorial models<sup>†</sup>

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>96,97</sup> and sustained interaction with adults<sup>98,99</sup> is known to be fundamental to early linguistic development. A review of three decades of literature<sup>100</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?

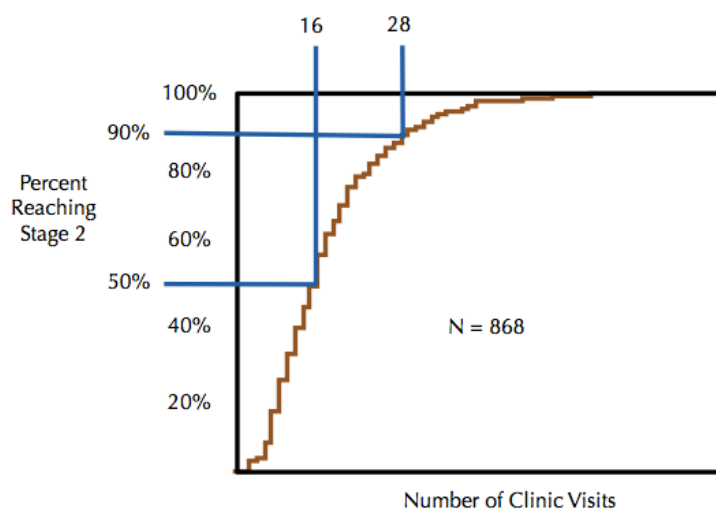
### The Lidcombe Program

There is a sufficiently comprehensive data set that gives an indication of how many Stage 1 clinic visits may be required with the Lidcombe Program; in other words, how many clinic visits are required to

<sup>†</sup> Thanks to Juliet Imeson for guidance with material in this section.

<sup>†</sup> Thanks to Michelle Donaghy for guidance with material in this section.

attain no stuttering or nearly no stuttering. More than a thousand children have been participants in Lidcombe Program clinical research, and the graph contains information about treatment time for 868 children, based on five file audits,<sup>56,66,101,102,103</sup> four clinical trials,<sup>4,9,14,31</sup> one prospective follow-up,<sup>56</sup> one translational study,<sup>45</sup> and one prospective observation study.<sup>68</sup> According to those studies, a median of 16 clinic visits is required for children to attain Stage 2 criteria, with a range of 11–23 clinic visits.<sup>†</sup> A file audit of 21 children treated by students under supervision in a university clinic<sup>104</sup> reported a median of 19 clinic visits to complete Stage 1. However 28 of the original 59 children in the study (57%) withdrew before completing Stage 1.



The range of median clinic visits to attain Stage 2 in the reports above is substantive, at 11–23. The graph<sup>‡</sup> is known as a recovery plot, and it shows the proportion of children to attain Stage 2 and the number of clinic visits to do so. With the median number of clinic visits being 16, half of those 730 children required fewer than 15 clinic visits for Stage 1, and half required more than 15 clinic visits. The estimated 90<sup>th</sup> percentile for the recovery plot is 28 clinic visits. In other words, 90% of cases will have attained Stage 2 by 28 clinic visits.

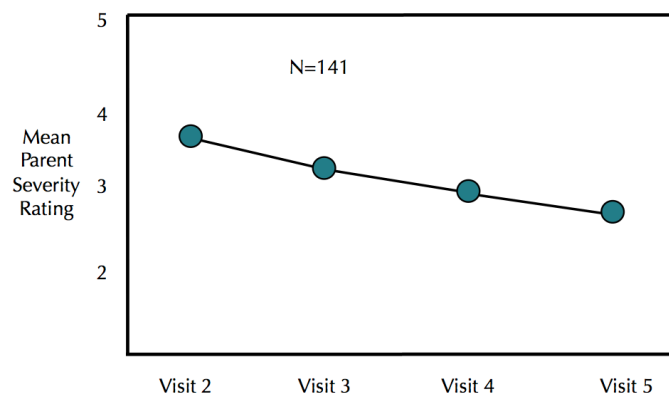
An important note here is that these data describe population caseloads of children, as do the results of clinical trials described earlier. As such, care is needed during the evidence-based reasoning cycle to form a judgement about the extent to which these data apply to any individual clinical child. One consideration will be comorbid diagnoses and case features. Two of the reports,<sup>101,102</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 comorbid with stuttering.<sup>105</sup>

<sup>†</sup> Two of those 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. Those reports contain a median of 11 clinic visits, compared to 16 for the remaining studies. Consequently, the treatment times in those reports were adjusted by adding five more clinic visits to their median values, bringing them into line with the subsequent studies.

<sup>‡</sup> 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.

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 graph<sup>‡</sup> is the mean parent SR for a report of 141 cases,<sup>103</sup> showing an average reduction of around one third during the first five clinic visits, or 4 weeks of treatment.



## HOW DOES A TREATMENT DELAY AFFECT THE TREATMENT PROCESS?

### The Lidcombe Program

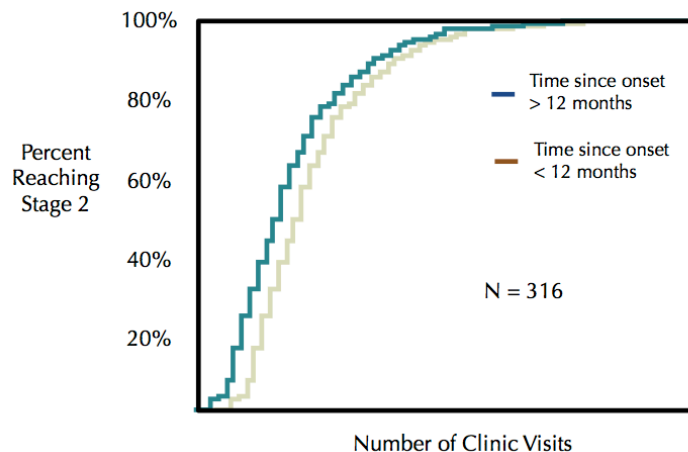
The next graph<sup>‡</sup> shows the results for 316 children in the studies mentioned earlier. The recovery plot on the left shows the children who had been stuttering for more than 12 months, and the recovery plot on the right shows the children who had been stuttering for less than 12 months. The children who had been stuttering for less than 12 months have the same shape of recovery plot as the others, 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 visits to reach Stage 2.

So it appears that delaying the Lidcombe Program for up to a year after stuttering onset is unlikely to jeopardise a child's responsiveness to it in terms of time taken to reach Stage 2.

That research result is intuitive, considering that the Lidcombe Program places a cognitive load on children to understand the treatment process. It appears from those 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.

<sup>‡</sup> 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.

<sup>‡</sup> 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.



## DO CASE VARIABLES AFFECT THE TREATMENT PROCESS?

### The Lidcombe Program

#### *Pre-treatment stuttering severity*

Six studies<sup>4,45,56,66,101,102</sup> have used a statistical technique called logistical regression to predict treatment time with the Lidcombe Program. Those analyses show that pre-treatment stuttering severity accounts for around 20% of the number of Stage 1 clinic visits required. That 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>68</sup> reported the same effect with a strong association. The study of the Lidcombe Program in a student clinic mentioned earlier<sup>104</sup> also reported that effect.

Another clinically useful perspective on this matter was presented in the Lidcombe Program translation report with community clinicians.<sup>45</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, that report found that receptive language scores and language development scores of Mean Length of Utterance, together, predicted 35–45% of the variance of clinic visits needed for Stage 1.

That 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 it in fact is not true.

## PARENT EXPERIENCES

### The Lidcombe Program

#### *The importance of this topic*

There have been three studies of how parents experience the Lidcombe Program.<sup>106,107,108</sup> Those reports provide useful information to forewarn clinicians about positive and negative features of the treatment that parents might encounter. In particular, clinicians can be forewarned about potential adverse parent experiences.

The first report<sup>106</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 reported lacking confidence to do it, but valuing 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).

The second report<sup>107</sup> found three typical paths through the treatment based on interviews with 14 parents of pre-schoolers being treated. Six of the parents were interviewed on two occasions. All but one interview was with mothers.

#### *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.

#### *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 for them. These parents became needy of support and the visits to the clinic became a burden for them, and they found the treatment difficult to sustain. Parents of these children began to be unresponsive and even irritated by parent verbal contingencies.

#### *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 in doing the treatment. These parents would doubt their capacity to do the treatment, and they focused 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>7</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*

The third report<sup>108</sup> involved 16 mothers who were each interviewed nine times during the course of 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, a number of 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 were 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 also there was an issue of just not expecting it to work:<sup>107</sup>

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)

Parents could be taken by surprise also by not expecting 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 children's reaction to the treatment. It was commonly reported that they were becoming aware of when they were stuttering and 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 like "great talking."

*Parent emotions*

Parents were reported to experience nine emotions during treatment. The five most common were judged as those 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 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, and guilt they were not doing the treatment correctly and 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 STUTTERING INTERVENTION EVIDENCE BASE: SUMMARY AND CONCLUSIONS**

### **The Lidcombe Program**

Independent reviews consistently report that the evidence base for the Lidcombe Program is the most comprehensive available among early stuttering treatments.<sup>109,110,111,112,113,114,115,116</sup> The latter review for 2015 lists 22 quantitative, data based research publications about the Lidcombe Program in peer-reviewed journals. That evidence base includes clinical trials, randomised clinical experiments, case studies, treatment process reports, and qualitative studies of parent experiences. Research publications continue to emerge, as described earlier during this lecture.

A strength of that evidence base is that it involves direct assessment of treatment effect size from randomised control trials and randomised controlled experiments that compared the treatment to 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>31</sup> That being 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 mechanisms that underlie the Lidcombe Program treatment effects. That research continues today, focusing on acoustic and linguistic variables as well as verbal contingencies. At present, the mechanisms underlying the Lidcombe Program remain unclear, but their eventual discovery may lead to development of a treatment with much different characteristics to 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>31</sup> although positive clinical trial results for this style of treatment await replication. This is a substantive issue 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 to three-quarters of the children in the randomised trial. It has also 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 to a no-treatment control group.<sup>43</sup> But as noted earlier, the RESTART trial provides no evidence that effect sizes, in terms of percentage stuttering reduction, are different from those obtained with the Lidcombe Program.

### **The Westmead Program**

This treatment is at the early stages of its development. Currently there is no randomised controlled evidence for it, and the only data available are nonrandomised Phase I and Phase II trials. In order to be as compelling as available evidence for the Lidcombe Program and RESTART-DCM Treatment, evidence from a randomised trial will be required.

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## LECTURE EIGHT

### EVIDENCE-BASED ADULT SPEECH TREATMENTS

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#### SPEECH RESTRUCTURING TREATMENT

##### Background

###### *Overview*

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> 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</sup> confirm that this approach is desirable for a substantive portion 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*, *fluency shaping*, 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*.

###### *Their mechanism*

These speech patterns 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, and increased voice onset time, vowel duration, and intervocalic interval.<sup>4,5,6,7,8,9</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 an inherently unstable speech motor system. If that is so, it is plausible that these speech patterns somehow, in a manner currently unknown, offset that problem by stabilising the speech motor system.

###### *A historical variant*

The terms *stutter more fluently* and *stuttering modification* are related to speech restructuring. Although they appear to be used less commonly these days, they refer to an alternative approach to control stuttering with a novel speech pattern. Charles Van Riper was an extremely influential clinician, largely because he developed the *stutter more fluently* technique.<sup>10</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. Terms for this technique are *cancellations*, *preparatory sets*, and *pull-outs*. Those techniques, and the history of their development are described in a reference text (p. 350–353).<sup>11</sup> However, as noted at that source,

“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. 353)

Since that statement, there have been two reports of treatments that have incorporated Van Riper’s techniques,<sup>12,13</sup> although neither report conformed to the definition of a clinical trial provided in Lecture Five. The former had all the features of a clinical trial except that speech measures were made in the clinic at 6 months post-treatment, and the latter was a file audit.

*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*. There seems to be some overlap with its use to reduce struggle with stuttering moments, as with *stutter more fluently* and *stuttering modification*, but it is also intended “to reduce fear, anxiety, and/or negative emotions associated with stuttering” (p. 1).<sup>14</sup> As such, it is classifiable within the class of anxiety management strategies known as *behavioural experiments*, which are discussed during Lecture Eleven. The report just mentioned<sup>14</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. 5).

**A brief history***The first records*

The long history of speech restructuring has been described in detail.<sup>15</sup> Lecture One described 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 the disorder. 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 obviously with conceptual similarity to modern speech restructuring:<sup>16</sup>

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. (p. 460)

It is clear that the use of speech restructuring variants occurred many times during subsequent centuries, and a full historical account is given in a 1984 text.<sup>17</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>18</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 the disorder.

*The 1960s onward ...*

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>19</sup> and a revolution began with treatment for chronic 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 systematically increased.

In the reference text mentioned earlier<sup>17</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 others.<sup>20</sup> Things have not changed since, with it being the most promising way for adults to control their stuttering should they wish to do so.

## PROGRAMMED INSTRUCTION

### A technique for behavioural control

More often than not, speech restructuring treatments reported worldwide incorporate programmed instruction, which is a technique invented by Skinner. Its principles and their application to speech-language pathology have been outlined in detail.<sup>21</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 predetermined 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, in the event of a stuttering moment occurring while a client is attempting to complete a step in the programmed instruction hierarchy, the client has to return 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 they are not substantiated by any research.

### Models of programmed instruction

Most speech restructuring clinical trials have involved programmed instruction, in a set sequence. Commonly, intensive treatment formats are used, which raises health economics issues about efficient use of clinical resources. For example, there has been an observation<sup>22</sup> that the results of a 5-day, non-residential intensive treatment<sup>23</sup> appear similar to the results of a 3-week residential treatment.<sup>24</sup>

In fact, intensive speech restructuring treatment does not seem essential to a positive treatment outcome. A clinical trial<sup>25</sup> suggested that 10 hours of individual treatment produced equivalent results to a treatment version involving an intensive treatment day. And there has been a clinical trial<sup>26</sup> showing that 16 two-hour sessions over four consecutive days produced equivalent results to two 2-hour sessions for 8 weeks.

### An example of programmed instruction

#### *Steps, targets, ranges and tasks*

With many programmed instruction stuttering treatments, the hierarchical sequence involves speech rate increments, as does the example in the table that uses speech rate in syllables per minute (SPM). Target speech rate increases with a sequence of six steps from extremely slow at 50 SPM, to a target speech rate of 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, of plus or minus 20 SPM in this case. For this example, there are nine speaking tasks within each step. The speaking tasks might involve monologues, or conversation with a clinician.

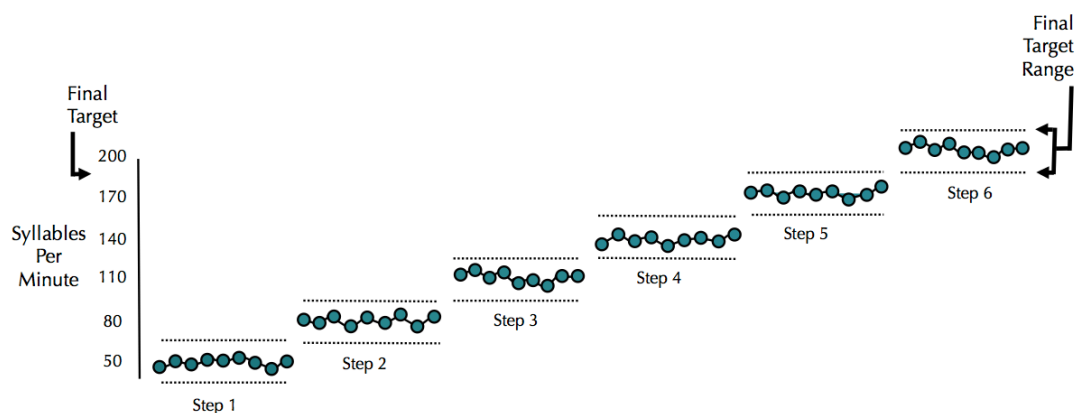


STEP	CRITERION ONE	CRITERION TWO	TARGET RANGE	SPEAKING TASK	TASKS PER STEP
1	50 SPM	No stuttering	+/- 20 SPM	200 syllables	9
2	80 SPM	No stuttering	+/- 20 SPM	350 syllables	9
3	110 SPM	No stuttering	+/- 20 SPM	500 syllables	9
4	140 SPM	No stuttering	+/- 20 SPM	650 syllables	9
5	170 SPM	No stuttering	+/- 20 SPM	800 syllables	9
6	200 SPM	No stuttering	+/- 20 SPM	950 syllables	9

The hierarchical sequence does not need to be speech rate as with this example. Many treatment programs do use speech rate with programmed instruction, such as this example.<sup>24</sup> However, the sequence can be increments of speech naturalness using the scale described during Lecture Four,<sup>27</sup> or it can be increments of utterance length using words or time.<sup>28,29</sup>

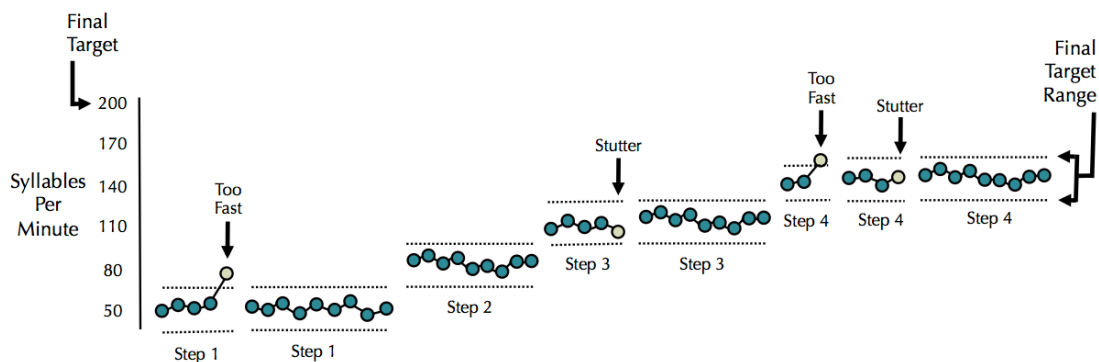
#### *The programmed instruction treatment process*

Here is a schematic diagram of a client progressing through a programmed instruction sequence such as in the table. The client progressed through the programmed instruction sequence without failing to meet stuttering or speech rate criteria during any speaking task.



However, the next diagram shows a likely scenario for a client who finds the programmed instruction sequence challenging and progresses through it with some contingencies for not meeting criteria. In theory at least, this treatment process would be of more use to a client because it involves some learning along the way about stuttering control.

During the fifth speaking task of Step 1, the client spoke too fast, and hence had to re-start the sequence of nine speaking tasks. Step 1 was completed at the second attempt. Step 2 was completed successfully at the first attempt. However, during the fifth speaking task of Step 3 a stuttering moment occurred, and the client re-attempted Step 3. This second attempt was successful. The first attempt at Step 4 failed because the client spoke too fast, and the second attempt failed because of a stuttering moment. A third attempt at Step 4 was successful, and the client is ready to attempt Step 5.



### 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 have the newly learned speech skills generalise 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

### Numbers of trials

There have been more positive clinical trials of speech restructuring for adults than for any other treatment. Using the definition of a clinical trial presented during Lecture Five, a total of 24 clinical trials have been published for adults:

Phase I: <sup>30 31 32 33 34 35 36 37 38 39</sup>  
 Phase II: <sup>23 24 26 27 40 41 42 43 44 45 46 47</sup>  
 Phase III: <sup>25 48</sup>

Those trials are by many independent researchers in different countries, dating from 1973. That being said, the same research group conducted the only two randomised Phase III trials.

### Effect size

As discussed during Lecture Five, effect size for a treatment can be assessed by comparing a treatment arm in a clinical trial to a control arm that receives no treatment. However, neither of the randomised trials of speech restructuring 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>27</sup> 20 of 32 dropped out, in another,<sup>24</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>43</sup> one with 39 participants,<sup>42</sup> and another with 44 participants.<sup>47</sup> In fact, only one trial with substantial numbers<sup>23</sup> reported few drop-outs: two of 80 participants. So the usual bias where non-randomised trials overestimate effect size<sup>49</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>25,48</sup>

<sup>†</sup> That reasoning assumes drop-outs are connected to undesirable features of the treatment. However, it is possible that participants will drop-out from clinical trials because the treatment is so rapidly efficacious that they do not bother to continue their participation in the trial.

In light of all that, 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.

Again with the reservation that the clinical trials concerned are 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 showing sustained stuttering reductions were 9–12 years (N=12),<sup>50</sup> that being a long-term follow-up of a trial,<sup>27</sup> and 10 years (N=17).<sup>46</sup> Both results were for multi-week, intensive residential treatments.

As mentioned several times during these lectures, post-treatment relapse is a recurring problem with speech restructuring treatment.<sup>51,52</sup> Relapse rates were specifically reported in some of the clinical trials that have been published for adults. For example, one trial<sup>43</sup> reported 30–60% relapse at 12–18 months, depending on how relapse was defined, and another trial<sup>24</sup> reported 24% relapse in terms of more than 6.0 %SS at 12 months post-treatment.

### Speech naturalness

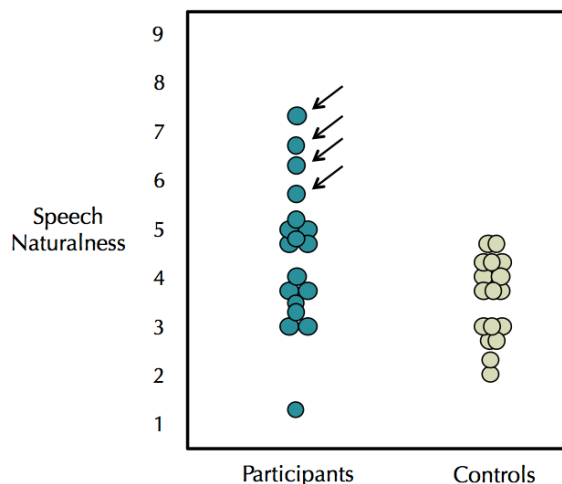
#### *How speech sounds*

As discussed during Lecture Four, gains from speech restructuring treatment are typically achieved at the cost of speech that does not sound perfectly natural. That problem has been known for decades.<sup>51</sup> There is an extensive body of literature dealing with the problem of post-treatment speech naturalness with speech restructuring treatment; 30 pertinent publications are documented in a reference text<sup>11</sup> (p. 341–342). 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>53</sup> A theoretical mechanism for that would be that unnatural sounding speech prompts listeners to evaluate such speech negatively, causing anxiety for the speaker. Such anxiety is known to have a detrimental effect on maintenance of speech restructuring treatment benefits, as described during Lecture Ten.

This is a recent statement of the problem of speech naturalness and speech restructuring treatment:<sup>54</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)

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<sup>‡</sup> from a clinical trial<sup>40</sup> illustrates the issue. Listeners assigned speech naturalness (NAT) scores 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 graph shows that, with the exception of four participants marked with arrows, the group who received treatment attained NAT scores around the range of controls.



<sup>‡</sup> 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.

*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>55,56</sup> The latter report 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 replaced it with a different feeling of being different: feeling different from their normal 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 I: THE CAMPERDOWN PROGRAM****Background***Overview*

This treatment is an example of a non-programmed speech restructuring model for adults. In summary, the treatment incorporates a video demonstration of the Camperdown Program *Training Model*. The clinician guides the client in using the speech pattern in that Training Model to develop an individualised *fluency technique* to reduce or eliminate stuttering during everyday speech and to sound as natural as possible.

*Nonprogrammed instruction*

The development of the Camperdown Program as a nonprogrammed treatment was prompted by a laboratory experiment<sup>57</sup> with three adults who had never experienced speech restructuring treatment. They learned a speech restructuring pattern and then were able to use it to control stuttering and sound reasonably natural simply by being instructed to do so. Programmed instruction was not necessary for them to attain that laboratory result.

*No speech targets*

As described earlier, speech restructuring clinical procedures typically involve teaching clients target speech behaviours: extended vowel production, light articulatory contacts, gradual onset of vocalisation, and continuous breath flow during speech. However, those speech targets have long been recognised as a threat to treatment replicability.<sup>58</sup> The term treatment replicability refers to whether clinicians can do a treatment in the same way as the clinicians did the treatment in published clinical trials.

The replicability problem with such speech targets was illustrated with a report<sup>59</sup> where seven clinicians with experience with one particular speech restructuring treatment were shown video recordings of clients demonstrating the target speech pattern at various stages of their treatment. The clinicians did not at all agree about whether the clients were using the program target behaviours correctly or incorrectly.

That result prompted the Camperdown Program to be developed without using any speech targets during the treatment process. Instead, the clinician shows the Training Model of the required speech pattern to clients and asks them to imitate it and to control stuttering while doing so. The clinician gives feedback about how closely the client imitates the Training Model, using whatever instruction is appropriate.

*No transfer phase*

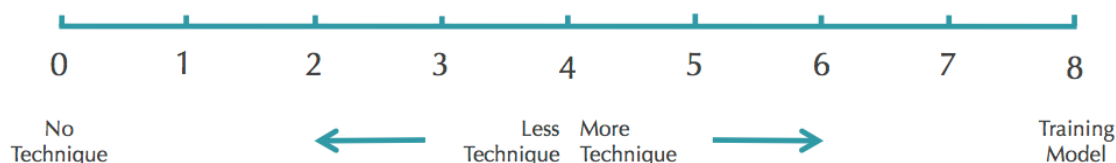
Another report<sup>31</sup> that influenced the Camperdown Program development showed that a traditional speech restructuring treatment involving a transfer phase remained efficacious when its transfer phase of treatment was replaced with speech practice. In response to that publication, Camperdown Program development proceeded without a formal transfer phase.

### Resource materials

The Camperdown Program Treatment Guide is downloadable from the Australian Stuttering Research Centre website.<sup>60</sup> Additionally, at that website there are downloadable video demonstrations of the Camperdown Program Training Model at around 70 SPM. The website also contains other downloadable clinical materials for use during the treatment process. The following description of the Camperdown Program draws freely from the treatment guide. A publication<sup>61</sup> outlines the use of technology during the treatment process, such as the Scenari-Aid website.<sup>62</sup>

### Stage I: Teaching treatment components

Stage One typically involves weekly clinic visits of around 1 hour duration. One purpose of Stage One is for clients to learn imitation of the Camperdown Program Training Model using one of the downloadable video examples. Another purpose of Stage One is for clients to learn how to use the Stuttering Severity Scale and the Fluency Technique Scale. The Stuttering Severity Scale has nine points, where 0 = *no stuttering*, 1 = *extremely mild stuttering*, and 8 = *extremely severe stuttering* (see Lecture Four). The Fluency Technique Scale was based on research that developed the scale of speech naturalness described during Lecture Four. It has nine points, where 0 = *no technique* and 8 = *very obvious technique*. The use of these scales is described in detail in the Camperdown Program Treatment Guide. The Fluency Technique Scale is below.



Clients are required to imitate the Training Model using fluency technique 7–8 and to speak using it spontaneously without stuttering, with a stuttering severity score of 0. Clients imitate the video Training Model and the clinician gives feedback about the attempts without reference to any speech targets. The clinician may direct clients' attention to certain parts of the Training Model and encourage them to listen again and to try to copy that section more closely.

Clients learn to use the Stuttering Severity Scale by giving a score to their speech from recordings and in real time for short periods of 1–5 minutes. Clients compare their stuttering severity scores with those of the clinician. Clients are required to measure their severity to within one scale value of the clinician's score.

### Stage II: Establishing stutter-free speech

During Stage II clients:

- (1) Consolidate their learning of the Training Model from Stage I
- (2) Work with the clinician to develop an individualised fluency technique that sounds natural and useable for stuttering control
- (3) Continue self-evaluation of their stuttering severity and fluency technique
- (4) Establish problem-solving skills for Stage III, which involves generalisation of stutter-free speech to everyday speaking situations

Stage II can be done with weekly clinic visits of around an hour duration, or with an intensive group format. Typically, clinicians find the former to be a more practical option.

### Fluency cycles: Overview

During Stage II, clients rotate through a series of fluency cycles. They use massed practice for clients to establish their own fluency technique that works to control their stuttering and, eventually, sounds as

natural as possible. One cycle consists of three parts: *Fluency Technique Practice*, *Experimentation* and *Planning*. Each part takes 3–4 minutes. Clients complete as many of these cycles as needed to achieve program criteria and progress to Stage III.

#### *Fluency cycles: I. Fluency Technique Practice*

The aim of Fluency Technique Practice is for clients to consolidate their learning of the Training Model that occurred during Stage I. Repeating this consolidation process during a series of “fluency cycles” ensures that the basic skill of controlling stuttering is continually reinforced. Clients do not make any attempt to sound natural. Client practise the Training Model at fluency technique 7–8. Throughout Fluency Technique Practice, the aim is for clients’ speech to continue to sound like the model and to remain stutter-free. After practising, clients record a stuttering severity score and a fluency technique score. The clinician gives feedback in the same manner as during Stage I.

#### *Fluency cycles: II. Experimentation*

An Experimentation part follows the Fluency Technique Practice part of a fluency cycle. The aim here is for clients to develop an individualised fluency technique and begin to sound more natural while maintaining low levels of stuttering. Clients experiment with using and evaluating as many different features of the Training Model as might be needed to control stuttering. It is intended that each client will ultimately develop an individualised fluency technique to control stuttering.

The Experimentation part consists of three steps: *Goal setting*, *Evaluation after speaking*, and *Evaluation after listening to the speech recording*.

*Goal setting.* The client sets stuttering severity and fluency technique goals to achieve. The primary goal is to remain stutter-free, hence the stuttering severity score goal is always 0. However, clients decide the fluency technique goal, differently each time if necessary. Clients determine the fluency technique goal by:

- (1) Reviewing evaluations of previous cycles in terms of their success with controlling stuttering
- (2) Evaluating how much fluency technique will be needed to control stuttering.

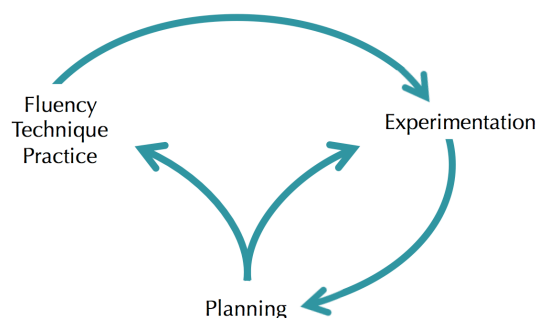
It is important for the client to do this goal setting with limited guidance from the clinician. Being able to plan a stuttering control strategy based on evaluation of previous performance is critical to the success of the Camperdown Program. The client then speaks for 3–4 minutes attempting to meet the set goals. The client makes an audio recording of this speaking task.

*Live evaluation.* The second step is client evaluation of the speaking task without listening to the audio recording. Clients record a stuttering severity score and fluency technique score for the speaking task. The clinician does not discuss these scores with the client at this stage. This process simulates everyday situations where clients need to evaluate their speech, and make decisions about stuttering control without clinician assistance.

*Recording evaluation.* The third step is evaluation of the audio recording. The client listens to the audio speech recording to confirm or correct the evaluations made during evaluation after speaking. This is done in consultation with the clinician and agreement about the evaluation needs to be reached by both.

#### *Fluency cycles: III. Planning*

This is the third part of a fluency cycle. The aim is for clients, initially with guidance from the clinician and ultimately alone, to use self-evaluations from previous fluency cycles to plan a strategy and set stuttering severity and fluency technique goals for the next cycle. During the planning part of a fluency cycle there are two ways for clients to proceed, as overviewed in the diagram.



If the client attains a stuttering severity score of 2 or more during the previous Experimentation part of a fluency cycle, the client returns to the Fluency Technique Practice part to start the next cycle.

If the client attains a stuttering severity score of 0–1 during the previous Experimentation part of a fluency cycle, the client chooses to begin the next cycle either at Fluency Technique Practice or Experimentation. Clients who choose the latter may choose to speak with a more natural sounding fluency technique goal than the previous time.

Regardless of how clients proceed at the Planning part of a fluency cycle, they begin at least every third cycle with the Fluency Technique Practice part. The point of this is to constantly consolidate their basic fluency technique skill.

### **Stage III: Generalisation**

During Stage III clients attend the clinic each week for the following purposes:

- (1) Consolidate use of their fluency technique to control stuttering
- (2) Compare their speech measures during the visit with the clinician's
- (3) Review their fluency technique practice routine with the clinician and revise it as needed
- (4) Present recordings of their speech and speech measures in daily situations and discuss them with the clinician
- (5) Use those recordings and speech measures as a focus of discussion with the clinician to deal with any emerging problems
- (6) Devise a hierarchy of difficult speaking situations to assist generalisation
- (7) Modify their measurement procedures for the coming week if needed

Clients progress to Stage IV when stuttering and fluency technique goals are met for three consecutive weekly consultations

### **Stage IV: Maintenance of treatment gains**

The goals of Stage IV are for clients to:

- (1) Maintain target stutter-free speech during the clinic visit
- (2) Present target stuttering severity and fluency technique scores for typical speaking situations
- (3) Present audio recordings of themselves to confirm those scores
- (4) Discuss with the clinician how they have dealt with any problems sustaining treatment benefits

The client attends 1-hour clinic visits that become less frequent according to progress. Discharge occurs when the client and clinician are satisfied that the client has developed self-management skills that are sufficient to sustain treatment gains. Commonly, clients have access to local self-help group meetings, which may be helpful during Stage IV.

### **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 Phase I and Phase II trials of this speech restructuring treatment model for adults. Two involved a standard clinic treatment format<sup>32,40</sup> and one reported results at a university student clinic.<sup>35</sup> One was a low-tech telephone telepractice trial.<sup>33</sup> Another was an experimental version of a standalone Internet presentation of the treatment that did not require a clinician.<sup>36</sup> The results of that standalone Internet Phase I trial with two adults was encouraging.

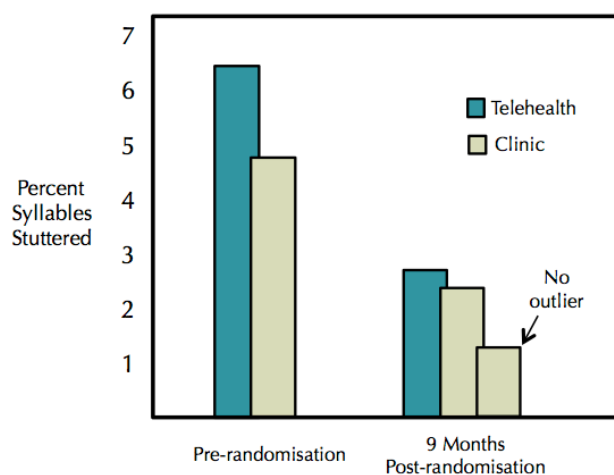
A subsequent trial of that standalone Internet version recruited 20 adults.<sup>37</sup> As with all clinician-free Internet treatment programs, compliance was an issue. Five participants completed the treatment and five completed more than half of it. Four of the five who completed the treatment reduced 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 the previous lecture.

#### *A Phase III clinical trial*

A randomised Phase III trial<sup>25</sup> compared the standard clinic Camperdown Program presentation with an experimental, low-tech telepractice version presented by telephone. For the telepractice adaptation, “home practice replaced the face-to-face programme group intensive day” (p. 110).<sup>25</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.

As mentioned during Lecture Six, there are some compelling advantages for telepractice treatment services with pre-schoolers. In the case of adults, there is the additional advantage for the many young adults who wish to reduce their stuttering for employment reasons, and are reluctant to take time off work for treatment. Additionally, treatment within the client home maximises cultural and community support during treatment.<sup>63</sup>



Results are presented in the figure.<sup>‡</sup> The telepractice 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 telepractice group did better considering that they were more severe pre-randomisation.

There is another reason to think that the telepractice group did better than the standard, in-clinic group. This is because of what is known as an outlier in that group. One of the in-clinic participants did not respond to the treatment at all. When the authors (controversially) removed that participant from the analysis the results for the telepractice group looked even better.

The telepractice 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 telepractice group used 221 minutes less contact time—3.7 hours—than the standard group. That result was clinically significant and also statistically significant.

Speech naturalness assessment used a control group as a reference. Results were similar to those for a Phase I trial of the Camperdown Program.<sup>40</sup> 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.

<sup>‡</sup> 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.



*No replications*

On balance, this body of clinical trial evidence might be interpreted as a sound data base attesting to 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 II: 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>64</sup> and is conducted at the Institute for Stuttering Treatment and Research<sup>65</sup> at Edmonton, Canada. It appears that a downloadable treatment manual is not available, however a description of the treatment as it is currently conducted is available.<sup>66</sup>

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>66</sup> That 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>66</sup>

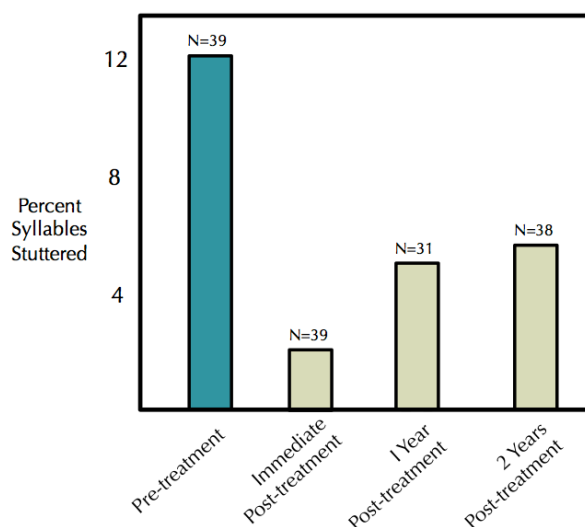
Those speech targets are taught with constant attention to attaining natural sounding speech, and clients learn to use a 10-point speech naturalness scale, which is used during the treatment process. The treatment incorporates Van Riper’s “stutter more fluently” techniques that were described earlier.

The Comprehensive Stuttering Program involves a substantive nonbehavioural component with cognitive behaviour therapy for each client (a psychological intervention: see Lecture Eleven). 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>24,30</sup> the former involving 42 participants, 22 of whom were adults. Subsequently, a Phase II trial was reported<sup>42</sup> involving 39 participants, 14 of who 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>42</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 that 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) 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).<sup>42</sup> This might be interpreted to suggest that Table 1 (p. 241) of the report shows unusually slow and hence unnatural sounding speech.

In another report with one of the longest follow-up periods on record,<sup>46</sup> this 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>42</sup> No speech naturalness data were presented to bolster confidence in that result. However, self-report data indicated that<sup>46</sup>

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)

## **SPEECH RESTRUCTURING III: 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 problem behaviours,<sup>67</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>68,69</sup> With stuttering, two single-subject laboratory experiments<sup>70,71</sup> showed promise for children and adolescents, with encouraging results continuing at 2–4 years follow-up.<sup>72</sup> Subsequently, a laboratory study<sup>73</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*

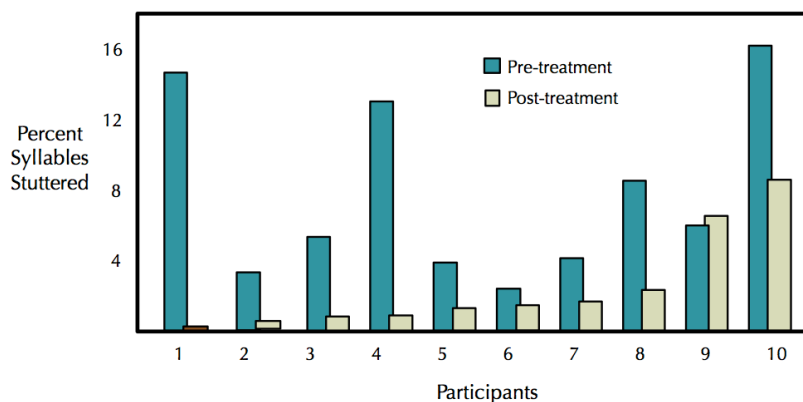
A case study report<sup>34</sup> explored the potential value of video self-modelling with the common relapse problem after speech restructuring stuttering treatment. The study recruited 12 adults who had

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,<sup>‡</sup> 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 to levels 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 with the result for another speech restructuring treatment by the same research group,<sup>40</sup> suggesting that regaining of stuttering control required some compromise of speech naturalness.



### Clinical implications

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>41,66,74,75,76</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 of whom were adults, were recruited into a Phase III trial.<sup>48</sup> One arm was standard speech restructuring and the experimental arm was standard speech restructuring plus video self-modelling. The participants were treated at different clinics around Australia, using different speech restructuring treatment models.

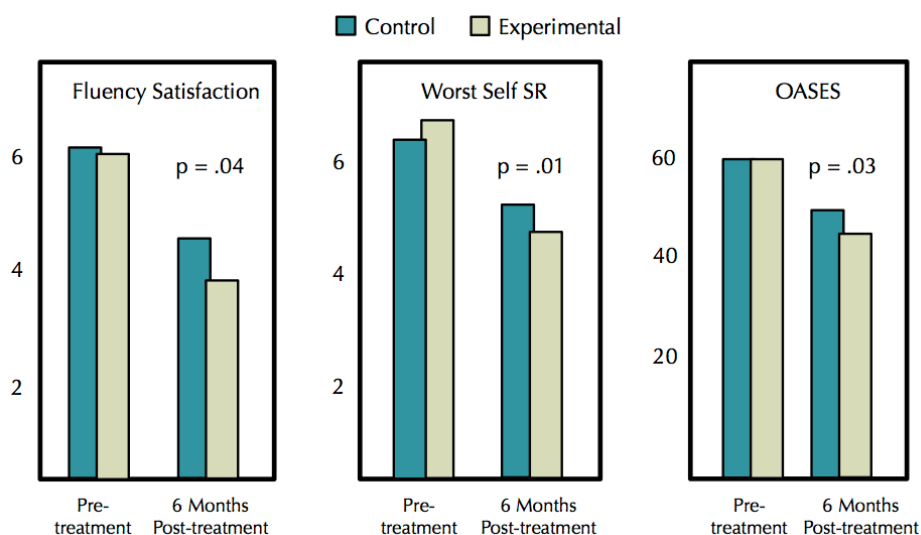
Towards the end of the treatment, the experimenters made video self-modelling tapes of all clients, lasting 4.5 minutes. Clients in the experimental group were given their self-modelling videos and instructed to watch them for 5 minutes each day for a month, with the following instruction: “Try to talk without stuttering using your speech technique as you see yourself doing on the video” (p. 890–891).<sup>48</sup> Five participants (6%) dropped out, and their results were analysed with intention to treat analysis by last observation carried forward.

<sup>‡</sup> 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.

The primary outcome was %SS measured from unscheduled telephone calls to participants from strangers. There were several secondary outcomes: Subjective Units of Distress<sup>77</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 quality of life measured with the Overall Assessment of the Speaker's Experience of Stuttering (OASES)<sup>78</sup> (see Lecture Four).

### 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 impairment at pre-treatment to mild-moderate impairment at post-treatment. Results are presented in the figure. 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, in the event of impending relapse, a client could return to daily watching. Only in the event that a return to daily watching failed 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>79</sup> were three adults who had received an intensive, residential speech restructuring treatment.<sup>80,81</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 experimenters 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. 9).

## **A VERBAL RESPONSE CONTINGENT TREATMENT: SELF-IMPOSED TIME-OUT**

### **The procedure**

Lecture One overviewed the extensive laboratory evidence of the controlling properties of response contingent stimulation, and Lecture Six outlined the Lidcombe Program model of verbal response contingent stimulation for treating early stuttering. A model of verbal response contingent stimulation suitable for adults that has supportive clinical trial evidence is known as self-imposed time-out. This means that when a stuttering moment occurs, the client stops speaking for a few moments, then resumes speaking. The duration of that period of self-imposed time-out seems not to matter.<sup>82</sup> All that seems to matter is that it is contingent on stuttering.<sup>83</sup> Research participants generally choose quite a brief period.<sup>84</sup>

### **Clinical advantages**

The advantage of the self-imposed time-out technique is that it does not use an overt speech pattern. Additionally, there is reason to believe that it might invoke existing speech skills, such as those learned with speech restructuring.<sup>85,86</sup> And in fact, three of four participants in a laboratory report<sup>85</sup> who had severe stuttering, and who attained more than a 60% stuttering reduction with self-imposed time-out, had received previous speech restructuring treatment.

### **Clinical trial evidence for self-imposed time-out**

#### *Phase I trials*

The first clinical trials of verbal response contingent stimulation with adults were Phase I trials<sup>87,88</sup> according to the Lecture Five definition. Those reports demonstrated potential value of the technique with single-subject experimental designs, each report using one participant.

#### *A randomised Phase II trial*

A treatment described as habit reversal<sup>89</sup> was developed originally to treat “nervous habits and tics” and later was applied to adult stuttering clients in a randomised Phase II clinical trial,<sup>90</sup> described as regulated breathing. The treatment is multidimensional, but appears to focus mostly on self-imposed time out:<sup>90</sup>

In order to regulate breathing, the client was instructed to stop speaking when a stuttering episode occurred and to take a deep breath by exhaling and then inhaling. (p. 41)

Forty adults were randomised to five treatment arms, four being a variant of the regulated breathing procedure and a fifth being a self monitoring placebo. It is difficult to interpret a five-arm trial with only eight participants per group, but modest stuttering reductions of around 50% were reported.

#### *A nonrandomised Phase II trial*

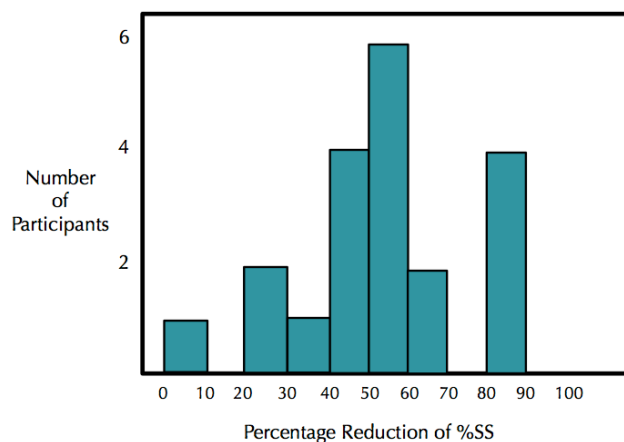
The last trial of self-imposed time out to date<sup>91</sup> was a non-randomised Phase II trial that recruited 30 participants, 26 of whom were adults. The trial involved instatement, generalisation and maintenance phases of treatment. Twenty-two participants completed the first two phases and 18 completed maintenance and remained in the trial at 6 months post-treatment. Therefore, the trial was affected by a high drop-out rate of 40%.

The instatement phase began with the clinician first imposing time-out and then the client learning to self-impose it. Subsequently, participants learned to use self-imposed time out during everyday speaking situations and to self evaluate their speaking performance. A subsequent maintenance phase assisted participants to sustain their treatment gains.

The mean reduction of %SS scores for the 22 participants completing the instatement and generalisation phases was 54% from pre-treatment to 1 week post completion of those phases. That result was sustained for the 18 participants for whom data were available at 6 months post completion of the instatement and generalisation phases.

As shown in the graph,<sup>‡</sup> more than half of the participants reduced their stuttering by more than 50%, and four of them reduced their stuttering by 80–90%. The report indicated that there was no difference in responsiveness between the adults and the four adolescents in the trial. The figure shows the percentage reductions for 22 participants at the conclusion of the instatement phase.

Speech naturalness for participants was assessed with the NAT scale, with the result that “around half of the participants achieved post-treatment speech naturalness scores within or near the range of control subjects” (p. 38).<sup>91</sup>



### *Clinical implications*

The verbal response contingent treatment model seems to have some treatment effects with adults and adolescents, but it seems not to have the strong effects that appear to occur for pre-schoolers. It is fairly clear that the clinical trial evidence for them is, on balance, neither as comprehensive nor compelling as for speech restructuring. However, it is arguable that the evidence is strong enough to suggest that they will have clinical value for some adults and adolescents.

The most obvious disadvantage of the treatment is its limited clinical trial evidence base. And, although self-imposed time-out clearly is less effortful for clients than speech restructuring, it is not clear exactly how much effort clients must use to sustain benefits during everyday speech. It is unknown at present, for example, what proportion of stuttering moments must receive time-out for a clinical effect and whether that needs to be sustained to retain the effect. The Phase II trial merely states “the majority of participants reported that they were using time-out only ‘sometimes’ 6 months after therapy” (p. 40).<sup>91</sup>

Based on available clinical trial evidence, a verbal response contingent treatment model might not routinely be the first treatment of choice for adults and adolescents. There are, however, some situations where a clinician might consider them the first treatment of choice. Examples include when a client is unable to learn a speech restructuring pattern, or cannot sound natural enough, or has a long history of unsuccessful treatment with speech restructuring, or simply does not like the speech restructuring technique.

## **MACHINE AIDED TREATMENTS**

### **Background**

#### *Potential benefits*

For obvious reasons, a machine that could do stuttering treatment automatically without a clinician, or produce clinically significant stuttering reductions when worn, would be well worth having, because

<sup>‡</sup> Adapted and reproduced with permission: Hewat, S et al (2001), Control of chronic stuttering with self-imposed time-out: preliminary outcome data, *Asia Pacific Journal of Speech, Language and Hearing*, 6, 97–102, © 2001 Taylor & Francis.

the clinician would have to do little, if anything. And if the machine was inexpensive and portable, clients could do their treatments when and where they chose. And if the machine reduced stuttering when it was worn, it would simply obviate the whole problem of chronic stuttering for the wearer during that period.

#### *A questionable history*

For those reasons, credible attempts to develop machine driven treatments are welcome. The history of clinical stuttering research contains many attempts to establish machine driven treatment,<sup>92</sup> and unfortunately, on many occasions those attempts have been accompanied by commercialism without sufficient evidence for clinical efficacy. The more memorable devices that could be worn by those who stutter are the electronic metronome and the Edinburgh Masker.

Both those 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. The 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 those devices was never determined by a clinical trial, and they appear to be no longer clinically available and not used much in clinical practice, if at all.

### **Altered auditory feedback**

#### *A topic of current interest*

The fluency enhancing condition called altered auditory feedback (see Lecture One) is delayed auditory feedback plus an alteration in pitch upwards or downwards. There has been much basic research about altered auditory feedback that has established a place for it as one of the fluency enhancing conditions. Around the same amount of basic research has been done about it as for time-out, although the effects of altered auditory feedback are not as reliable across participants as is the case for time-out. A recent laboratory study<sup>93</sup> reviews much of that literature.

#### *SpeechEasy*

Altered auditory feedback has emerged as a current controversial topic in stuttering research with the commercialisation and advertising of a device known as SpeechEasy.<sup>94</sup> It resembles a monaural hearing aid and presents altered auditory feedback to the wearer. One source of controversy about it emerged in a publication<sup>95</sup> in which the device was cited as an example of pseudoscience. Predictably, a vigorous exchange followed in the literature.<sup>96,97</sup> During the course of that exchange the developers of the device disclosed a financial interest in it.<sup>96</sup>

#### *A Phase I trial of SpeechEasy*

Altered auditory feedback clinical research has in common with time-out that the clinical trial evidence is not particularly encouraging. The only clinical trial that has been published is a Phase I trial of the SpeechEasy device by a group independent of its developers.<sup>98</sup>

Eleven adult participants were recruited and fitted with the device and wore it for 4 months, with a guideline to wear it for at least 5 hours per day. A little publicised feature of the device was documented in this trial. Namely, that it is not intended as a standalone device, but is intended to be combined with features of speech restructuring. Participants were instructed to use<sup>98</sup>

several active techniques to alter one's speech pattern, such as easy vocal onsets, prolongations, continuous phonation, starter sounds ... and fillers. Participants were told that these active strategies could be introduced at their discretion to help initiate voicing and/or enhance responsiveness ... (p. 520)

Participant speech was measured beyond the clinic on three occasions while conversing and asking questions of a stranger: pre-treatment, during the 4 months of wearing the device, and during a

withdrawal phase without the device. Results are summarised in the figure,<sup>‡</sup> with the means showing no effect from wearing the device. The error bars are the 95% confidence intervals.

#### *Differing interpretations of the trial*

The researchers who did the trial did not recommend moving on to a Phase II trial. The developers of the device protested vigorously,<sup>99</sup> claiming that the clinical trial was methodological flawed. The researchers who did the trial retorted by saying there was nothing wrong with their methods.<sup>100</sup>

#### *A subsequent report*

The developers of the SpeechEasy device then published a report<sup>101</sup> described as a “randomized clinical trial” with 18 adult Brazilian Portuguese participants diagnosed with stuttering. One group of 11 participants received SpeechEasy treatment for 6 months “with no training to use any fluency enhancing techniques” (p. 3). Another group of seven participants received treatment involving

both fluency shaping and stuttering modification techniques—negative practice, smooth speech, resisting time pressure and use of voluntary disfluencies. In addition to the practice of these speech motor skills, the treatment programme emphasized self-observation and included systematic cognitive and attitudinal intervention. (p. 3)

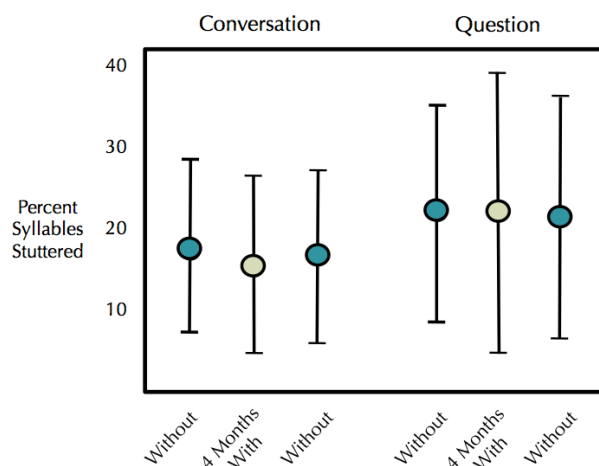
Pre-treatment and 6-months post-treatment assessments occurred, but the results of this report are uninterpretable because both groups showed only a 40% reduction of stuttering severity, and measures were based solely on 200 syllables of “monologue speech, conversational speech and oral reading” (p. 3) from within the clinic.

### **Modifying phonation intervals**

#### *The opposite of speech restructuring?*

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 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>102,103,104</sup> It is of interest that a report<sup>105</sup> showed four of seven clients who received a standard speech restructuring treatment reduced frequencies of short phonation intervals post-treatment. So, regardless of how successful modification of phonation intervals may prove to be as a treatment, it seems that modification of phonation intervals is not necessary for stuttering reduction.

The treatment phases incorporated programmed instruction requiring the client to speak without short phonation intervals. The machine fails participants at any step for not meeting phonation interval criteria. The modifying phonation intervals program contains instatement, generalisation and



<sup>‡</sup> Adapted and reproduced with permission: Pollard, R, et al (2009), Effects of the SpeechEasy on objective and perceived aspects of stuttering: A 6-month, Phase I clinical trial in naturalistic environments, *Journal of Speech, Language, and Hearing Research*, 52, 516–533, © 2009 American Speech-Language-Hearing Association.



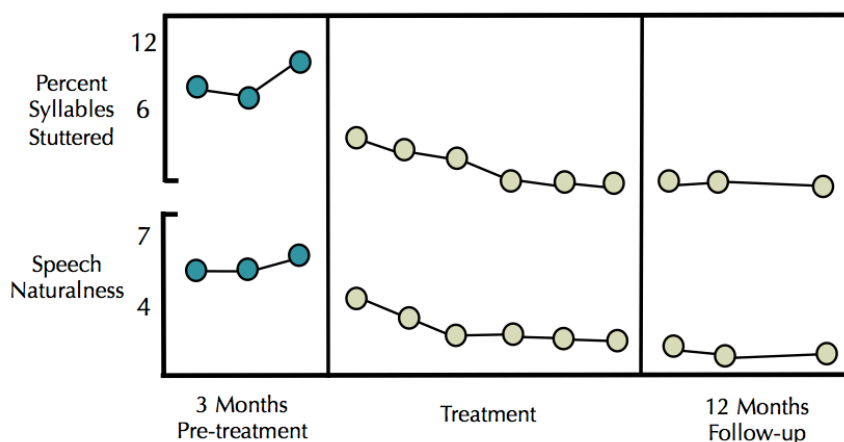
maintenance phases. The instatement phase is intensive, requiring six 3-hour treatment days per week, for a period of from 3–12 weeks, depending on the rate of client progress.

### Two data-based case studies

There have been no clinical trials published for modifying phonation intervals treatment. However, after the initial laboratory research findings, two data based case studies were published.<sup>106,107</sup> They are data based case studies rather than clinical trials because outcome measures within and beyond the clinic were a part of the treatment: 3-minute conversations for which the number of syllables was not reported. In other words, as part of their treatment, clients were trained to speak without stuttering in situations that were used to evaluate treatment outcome.

The first report<sup>106</sup> was five adult participants studied with a single subject experiment. The figure shows mean %SS and NAT scores for the five participants during a 3-month pre-treatment period. It also shows scores for the treatment period, which involved 2 weeks of instatement and 8 weeks of generalisation. Subsequently data are presented for a 12-month follow-up period. The figure shows one of several treatment data sets presented by the authors, based on telephone conversations within the clinic, without any feedback from the machine. Treatment speech measures beyond the clinic yielded similar results.

The figure<sup>‡</sup> shows that %SS and NAT scores were quite high during the pre-treatment period. The high NAT scores would be expected with the presence of clinically significant stuttering. The %SS scores reduced to clinically significant levels with almost no stuttering during treatment, and NAT scores stabilised at what appears to be within the normal range. Those results were sustained during a 12-month follow-up period.



The second data-based case study<sup>107</sup> compared 10 participants who received an intensive speech restructuring treatment<sup>27</sup> with 17 participants who received the modifying phonation intervals treatment. Surprisingly, five of the participants in this second report were participants in the first report.<sup>106</sup> Consequently, it is not possible to ascertain mean outcomes for the 12 novel participants in the second report. Additionally, it is not clear whether the MPI software and hardware were functionally similar in the two reports, or whether the five participants in the original report 14 years earlier used a different apparatus.

Regardless, the authors showed that there was no significant difference between the speech restructuring group and the modifying phonation intervals group at 12 months follow-up. It seems likely that the outcomes for the 12 novel participants in the second report attained similar outcomes to the five participants in the initial report, with the exception that they did not sound as natural according to scores with the 9-point NAT scale.

<sup>‡</sup> Adapted and reproduced with permission: Ingham, R J et al (2001), Evaluation of a stuttering treatment based on reduction of short phonation intervals, *Journal of Speech Language and Hearing Research*, 44, 1229–1244, © 2001 American Speech-Language-Hearing Association.

*Awaiting a clinical trial*

As yet, no clinical trial of the treatment has been published. The treatment is provided as a clinical service by certified United States clinicians who are trained at the University of California, Santa Barbara. The modification of phonation intervals software and hardware is available only for purchase by certified clinicians,<sup>108</sup> but is available as a smartphone application at another source.<sup>109</sup>

**Transcranial direct current stimulation**

This treatment development was spurred by applications of the technique to rehabilitation of limb and speech motor function with stroke patients, and a feasibility study with stuttering.<sup>110</sup> A randomised clinical experiment was reported with the technique as an adjunct to speech treatment for stuttering.<sup>111</sup> 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>111</sup>

Participants were randomised to receive five consecutive days of 20-minute speech treatments with transcranial direct current stimulation, and five consecutive days without that adjunct. 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, the authors concluded that “transcranial direct current stimulation combined with behavioural fluency intervention can improve fluency in adults who stutter” (p. 1).

**PHARMACOLOGICAL TREATMENTS***A long search*

Since the 1960s, there has been a search for a pharmacological stuttering treatment, and a large number of 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>112</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<sup>112</sup>

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)

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>113</sup>

A more recent review<sup>114</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, nobody seems to have thought of before: that drug treatment for stuttering might be improved if combined with standard, speech-language pathology interventions.

A subsequent report<sup>115</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 including 'air flow technique' and 'break Valsalva maneuver' as well as 'desensitization' from Van Riper's protocol" (p. S271). That, 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.

In short, at present there is no reason to consider drug treatment for stuttering, nor is there any reason to anticipate that there ever will be. 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.

## **SUMMARY**

According to clinical trials, speech restructuring is the most efficacious speech treatment for adults who stutter. Currently it is unknown how large the effect size is for speech restructuring treatments or for how many clients they will be viable. However, for some adults, the treatment clearly is capable of controlling stuttering to a clinically significant extent and for a clinically significant period. There is convincing evidence that video self-modelling is a useful adjunct to speech restructuring treatment. There is some clinical trial evidence that self-imposed time-out may be a treatment option in some circumstances. There is clinical trial evidence that the SpeechEasy device has no clinical value. However, there is promising Phase I evidence for another machine-based treatment: modification of phonation intervals. There is no reason to consider pharmacological stuttering treatments for clients.

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## LECTURE NINE

# EVIDENCE-BASED ADOLESCENT AND SCHOOL-AGE SPEECH TREATMENTS

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### SPEECH RESTRUCTURING I: INTENSIVE SMOOTH SPEECH

#### A Phase II trial

Smooth speech treatment is a variant of speech restructuring that involves<sup>1</sup>

instruction on respiratory control, where easy, relaxed diaphragmatic breathing during speech is demonstrated. ... another fundamental characteristic of smooth speech is the use of gentle onsets and offsets. This is achieved by starting the phrase with exaggerated airflow and using soft articulatory contacts. In addition, a phrase/pause speech pattern is taught. (p. 812)

The treatment incorporates programmed instruction with speech rate increments from 50 syllables per minute (SPM) and targeting a final rate of 160–200 SPM. There are formal transfer, generalisation and maintenance phases. The maintenance phase lasts 12 months.

A Phase II trial<sup>1</sup> had three treatment arms and a control group, although it was not randomised. Two of the arms were smooth speech, both given intensively, one with parents present and the other without parents present. The trial was for children 9–14 years, which spanned the ages of older school-age<sup>†</sup> children and younger adolescents (teenagers). It is the largest clinical trial for this age group in terms of scope and participant numbers.

For the smooth speech arm without parents present, participants received 5 hours of training prior to the treatment day to ensure that the requisite speech pattern had been learned. Then groups of 3–5 participants received intensive treatment for a week of 7-hour days. For the smooth speech arm with parents present, 2–5 parent-child pairs participated in 7-hour day groups which, were held once a week for 4 weeks. Parents were involved with the treatment process, and did treatment at home on non-clinic days.

Data based on recordings beyond the clinic were not available for the standard clinician group, but were available for the clinician-parents group. For that group, 25 children were recruited, with a mean age of 10.5 years, and beyond-clinic pre-treatment stuttering severity of 10.9 %SS. Twenty-seven children were recruited for the standard clinician group, with mean age 10.6 years. There were 20 children in the control group with mean age 10.9 years and pre-treatment stuttering severity of 8.8 %SS.

#### Results

It is not clear from the report whether there were any drop-outs. At 1 month post-treatment the mean stuttering severity in the clinician-parents group was 2.9 %SS. Control group data were not available at 12-months post-treatment. Speech rate increased for the clinician-parents group from 131 SPM pre-treatment to 174 SPM at 12 months post-treatment. To further assess speech naturalness the researchers argued that a 9-point scale was<sup>1</sup>

too difficult for the parent and child to make a distinct and easy judgment so it was adapted to a 5-point Likert scale, with 1 representing "poor," 2 "fair," 3 "moderate," 4 "good," and 5 "very good" speech naturalness. (p. 815)

Unlike the original 9-point scale described during Lecture Four, this scale was accompanied with a detailed description of how to use it. The mean speech naturalness score for the children was 2.6 at

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<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.

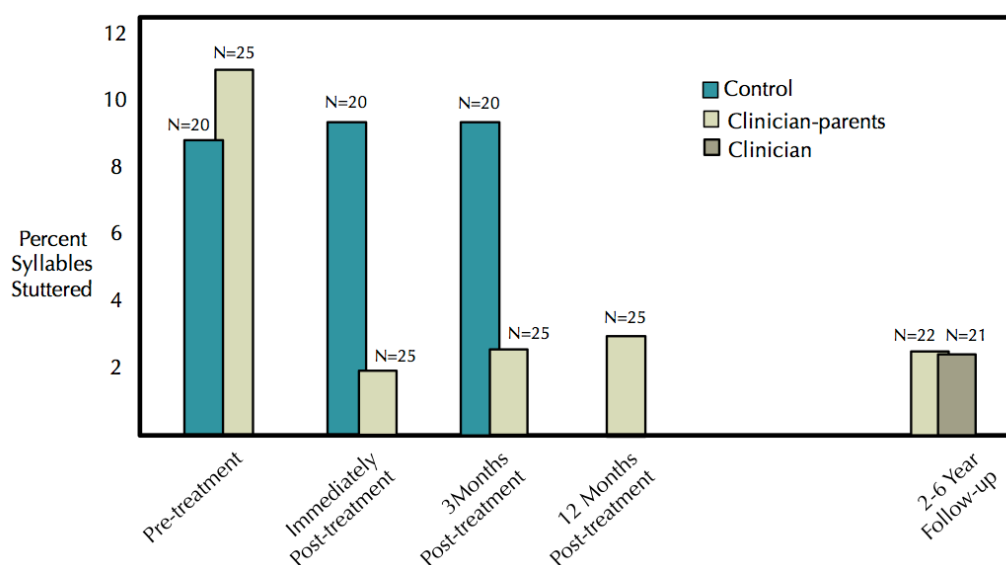
pre-treatment and 3.5 at 12 months post-treatment, suggesting that the children may have attained reasonably natural sounding speech. That result, however, is difficult to interpret because no control group was available at 12 months post-treatment, and no methodological details are presented for how the speech naturalness scores were obtained.

### Anxiety reduction

The trial used the State-Trait Anxiety Inventory for Children<sup>2</sup> as a secondary outcome. Anxiety for the treatment groups showed significant change from pre-treatment to 12 months post-treatment. However, it appears that those gains were not clinically significant, because the children were in the normal range of scores pre-treatment.

### Follow-up

A separate report for the trial was a 2–6 year follow-up<sup>3</sup> of 22 children in the clinician-parents group and 21 children in the clinician group. Treatment gains seemed to be retained in terms of %SS scores, with even faster speech rates than the original report, and better mean speech naturalness scores of 4.5. Results of the initial trial and follow-up are shown in the figure,<sup>†</sup> with mean %SS scores for beyond-clinic assessments.



## SPEECH RESTRUCTURING II: THE COMPREHENSIVE STUTTERING PROGRAM

### A Phase II trial

For this age group, the next most comprehensive Phase II clinical trial included 17 adults and 23 adolescents<sup>4</sup> and used the 3-week intensive Comprehensive Stuttering Program mentioned during the previous lecture. The adolescents were 12–17 years. Two children in the trial were 11 years, although the trial lists them as adolescents.

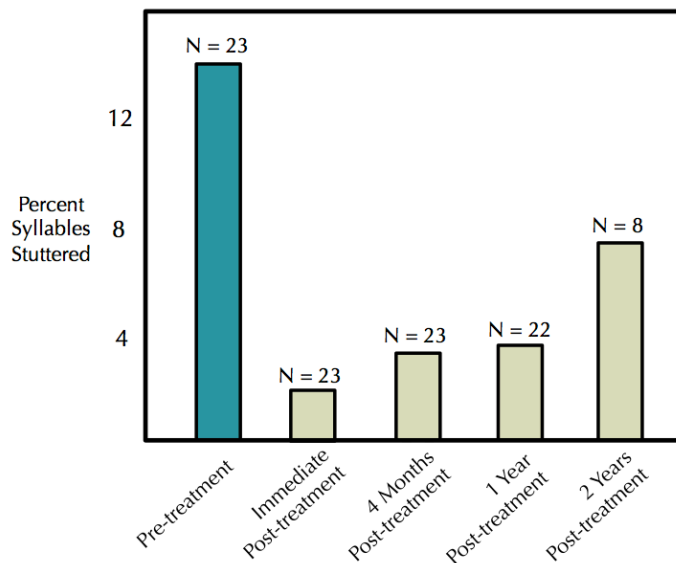
<sup>†</sup> Adapted and reproduced with permission: Craig, A et al (1996), A controlled clinical trial for stuttering in persons aged 9 to 14 years, *Journal of Speech and Hearing Research*, 39, 808–826, © 1998 American Speech-Language-Hearing Association.

## Results

Results for the adolescent participants, excluding the 11-year olds, were that stuttering severity remained just below 4 %SS at 1 year post-treatment, with some clinically significant regression at 2 years post-treatment. Results are presented in the graph. There was no indication that the adolescent participants responded differently to the treatment than the adults in the trial; both groups reduced around 75% of their stuttering.

At 1 year post-treatment there was only one drop-out, so in that sense the data are reasonably credible. Another credible feature of the data is that the 4 month and 2-year follow up %SS scores were obtained by telephoning clients unexpectedly when they had been discharged from the treatment facility.

However, it is nearly impossible to interpret the clinical value of these results because, as the authors acknowledge, no speech naturalness data or speech rate data were reported. During assessment, participants could have been speaking with unnatural sounding speech that had little functional clinical value during everyday life outside the periods of assessment.



The pre-treatment scores for the two 11-year old school-age children in the trial were 22.5 %SS and 13.0 %SS and their corresponding 1-year post-treatment scores were 0.8 %SS and 0.2 %SS. Again, it is difficult to interpret these results without any speech naturalness data.

To that result might be added that a clinical trial of a similar adult speech restructuring treatment, with a 2-week intensive format<sup>5</sup> included a 10-year old boy. His stuttering severity beyond the clinic was 5–10 %SS pre-treatment, and was at near-zero levels 12-months post-treatment. Speech naturalness, using the 9-point NAT scale, was 1.3 at 12 months post-treatment, suggesting natural sounding speech. A long-term follow-up of that clinical trial<sup>6</sup> reported that this boy, 9 years later, retained near-zero %SS scores beyond the clinic.

## SPEECH RESTRUCTURING III: THE CAMPERDOWN PROGRAM

### In-clinic

#### *A Phase I trial*

A Phase I trial<sup>7</sup> involved three adolescents who were treated with the Camperdown Program using a 1-day group format for Stage Two (instatement). Participants were three boys, ages 13, 14, and 16 years.

#### *Results*

One of the boys did not respond to the treatment at all and did not reach Stage Four (maintenance). Another boy withdrew from treatment during Stage Four but still showed a stuttering severity reduction of around half during unscheduled, beyond-clinic telephone calls. His pre-treatment score was 8.7 %SS and at 12 months follow-up was 4.2 %SS. He was, however, satisfied with his treatment outcome. The third boy did well with the treatment, with pre-treatment and follow-up scores of 4.2 %SS and 0.9 %SS, respectively. Results were confirmed with self-rated SR scores. Both boys attained natural sounding speech according to measures with the 9-point NAT scale.

## Telepractice

### *A Phase I trial*

A Phase I trial<sup>8</sup> involved telepractice treatment of three adolescents with the Camperdown Program using webcam: two boys, ages 13 and 15, and a 16-year-old girl. They were treated individually, with no clinic attendance at any time.

### *Results*

All participants entered Stage Four (maintenance). Participant 1 required 13 sessions over 16 weeks, requiring 8.3 hours of clinical time. Participant 2 required 24 sessions over 20 weeks, requiring 15.0 hours of clinical time, and Participant 3 required 16 sessions over 20 weeks, requiring 9.5 hours of clinical time. The mean times elapsed between webcam treatment sessions for each participant were 9, 6 and 10 days.

Naturally, with so few participants conclusions must be guarded, but there was some suggestion that the webcam telepractice format was a better prospect for these adolescents than those in the other Phase I trial<sup>7</sup> who were treated in a clinic. Results from unscheduled beyond-clinic telephone calls suggested that all participants improved. Pre-treatment and 12-months follow-up %SS scores were as follows: 16.7 to 8.4, 21.8 to 2.5, and 9.2 to 1.6. Results were confirmed with self-rated SR scores. All post-treatment speech samples fell within the range of speech naturalness scores attained by matched controls.

At another source describing this webcam trial,<sup>9</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 speech pathologist] than he is at the school he goes to, and with his teachers. This is a big winner."

### *Situation avoidance*

Situation avoidance—a marker of social anxiety (see Lecture Ten)—was measured with a 3-point scale, where 1 = *never avoided*, 2 = *sometimes avoided*, and 3 = *usually avoided*. Participants gave responses for each of eight common speaking situations, so total scores ranged from 8, indicating no avoidance, to 24, indicating maximum avoidance. The speaking situations were<sup>8</sup>

- (a) talking with a family member, (b) talking with best friend, (c) talking in a group of friends, (d) talking with an authority figure such as a teacher, (e) verbally providing name and address, (f) giving a class presentation, (g) talking on the telephone, and (h) ordering food or drink. (p. 374)

Two of the adolescent participants showed little clinical change of their pre-treatment avoidance scores at 12 months post-treatment. The other participant scored 19 at pre-treatment and 9 at 12 months post-treatment, suggesting a clinically significant improvement. In fact, that result suggests near-elimination of situation avoidance for that participant.

These results are particularly important; all three participants reduced their stuttering, but only one of them reduced situation avoidance scores to a clinically significant extent. In other words, reduction of stuttering was not necessarily associated with a reduction of anxiety. The two adolescent participants in this trial who did not reduce their situation avoidance would require a separate intervention to deal with that anxiety issue. Anxiety treatment for stuttering is considered during Lecture Eleven.

### *A Phase II trial*

A Phase II trial<sup>10</sup> from the same research group involved 16 boys, 15 of whom were adolescents, with mean age of 14 years 6 months, range 12–17 years. They were treated with similar webcam methods to the Phase I trial just described. Two participants dropped out from the trial.

### Results

There was a mean 55% stuttering reduction from pre-treatment to 12 months after entry to maintenance. The range of stuttering reduction was 0–100%. Two boys did not respond to the treatment and with them removed from the analysis the mean stuttering reduction to 12 months after entry to maintenance was 63%, with a range of 26–100%.

### Situation avoidance

Using the same situation avoidance measure as in the Phase I trial, there was a statistically significant reduction of scores from 15.3 pre-treatment to 10.3 at 12 months after entry to maintenance. However, not all the boys showed such a positive effect, with three of them persisting with clinically significant situation avoidance.

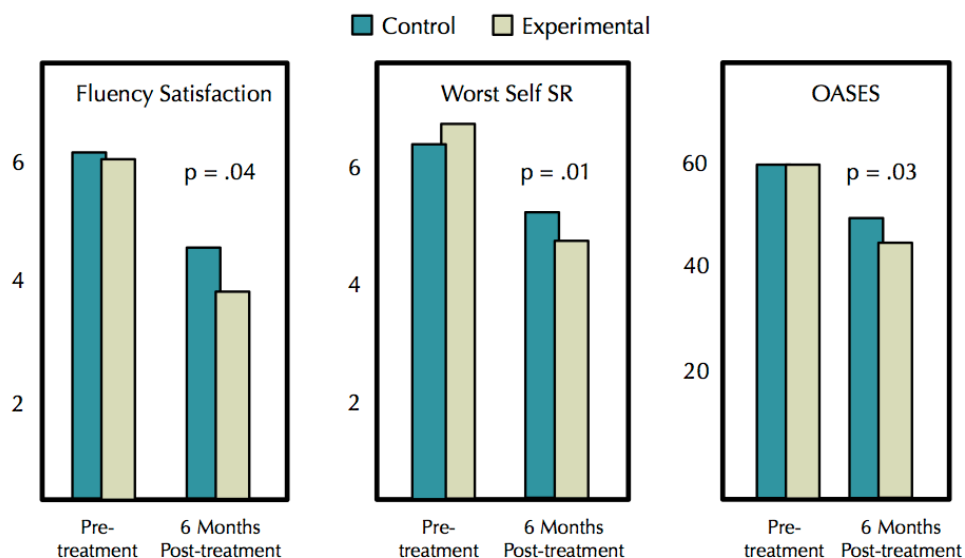
## SPEECH RESTRUCTURING IV: VIDEO SELF-MODELLING AS A SUPPLEMENT

### A Phase III trial

The trial of the video self-modelling technique outlined during the previous lecture<sup>11</sup> involved 25 participants who were adolescents, with a mean age of 13 years in the speech restructuring arm of the trial and 14 years for the experimental arm: standard speech restructuring plus video self-modelling. Eleven were randomised to the standard arm and 14 were randomised to the experimental arm. The report does not present data separately for adults and adolescents. However, the authors reported no statistical evidence of outcome differences between the adults and adolescents in the trial. Therefore, the findings for the adults in that trial summarised during the previous lecture might confidently be applied to adolescents.

### Results

To reiterate the results of the trial presented during the previous lecture, there were three significant findings: fluency satisfaction, improvement in the speaking situation self-rated most severely, and the OASES. The latter scores for the experimental group dropped from moderate impairment at pre-treatment to mild-moderate impairment at post-treatment. Results are presented in the figure.



## VERBAL RESPONSE CONTINGENT STIMULATION

### The Lidcombe Program

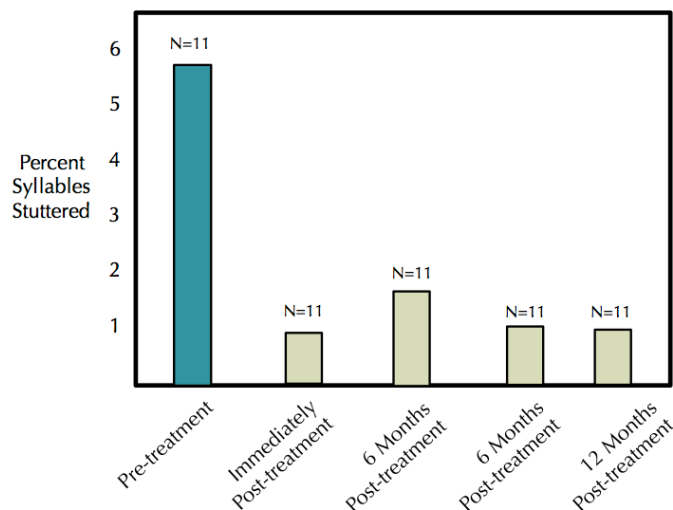
#### *A Phase II trial*

There has been one Phase II clinical trial of the Lidcombe Program for school-age children.<sup>12</sup> Fifteen children were recruited to the trial and four dropped out. The remaining children had a mean age of 8.3 years with a range of 6.10 years to 12.4 years.

#### *Results*

The trial showed a stuttering reduction of 89% from a mean pre-treatment stuttering severity of 5.3 %SS. A mean of 15 hours of treatment was required, with a median of 12 hours, to reach Stage 2. The data are presented in the figure. 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.

These results are consistent with a follow-up of eleven 6–10 year old children who were treated with the Lidcombe Program.<sup>13</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.



### Gradual Increase in Length and Complexity of Utterance (GILCU)

#### *Method*

Another model of verbal response contingent stimulation is Gradual Increase in Length and Complexity of Utterance, commonly referred to as GILCU. This well known treatment program has been detailed in two editions of a text.<sup>14,15</sup> The fundamentals of the procedure were first described in 1965<sup>16</sup> and variants of it have been described elsewhere.<sup>17,18,19</sup> The developer of the GILCU model spent many decades training clinicians to use it in the United States,<sup>20</sup> and it has been used in the United Kingdom,<sup>21</sup> Germany,<sup>22</sup> and Hong Kong.<sup>23</sup> GILCU has been discussed in several publications.<sup>20,24,25</sup>

The GILCU program begins with a programmed instruction Establishment phase containing many steps, beginning with the child being required to speak a word without stuttering and moving up through a series of requirements up to 5 minutes of reading, monologue, and then conversational speech. It qualifies as a model of verbal response contingent stimulation because the contingency for stutter-free speech is praise, sometimes paired with redeemable tokens. The prescribed verbal contingencies for stuttering are (surprisingly) “stop” and “speak fluently” (p. 64).<sup>18</sup>

Parents are trained to identify stuttering and to implement home practice. Branching steps are included to provide additional, remedial training when a child doesn’t succeed at any step. The procedure contains transfer and maintenance phases, during which verbal contingencies are systematically withdrawn.

#### *A Phase I trial*

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. The trial<sup>26</sup> recruited four children, one of whom dropped out. It is not clear from the report whether any of the children were adolescents. Probably, though, most were in the school-age range because those four children had a mean age of 11.3 years.

*Results*

Participants had a mean pre-treatment stuttering rate of 5.9 stuttered words per minute, and at 9 months follow-up the three that remained had a mean score of 1.0 stuttered words per minute, for an 83% stuttering reduction. For the Establishment phase, a mean of 9.6 hours of treatment was required, 6.4 hours for the Transfer phase, and 2.0 hours for the Maintenance phase. The report contained no speech naturalness assessments, perhaps with the assumption that speech naturalness would not be an issue with a GILCU procedure.

*Concluding comments*

A total of 208 participants of all ages have been reported in GILCU data-based clinical studies.<sup>25</sup> Yet only three of those participants in one report met a reasonable clinical trial requirement of beyond-clinic speech measurement with a follow-up period. It is therefore probably a reasonable observation that the GILCU clinical trial evidence base is not well advanced even though “the programme has been in use for 40 years” (p. 228).<sup>25</sup>

(One GILCU report<sup>18</sup> appears to be a clinical trial with a 14-month follow-up of 6 children. However, clinicians collected those data in the clinic. Beyond-clinic measures were obtained during the study, but the latest were at the end of the Transfer phase, without a follow-up period.)

**Self-imposed time-out***A Phase I trial*

The regulated breathing technique described during the previous lecture appears to focus mostly on self-imposed time out. Drawing on a clinical experiment with 21 school-age children and adolescents who stuttered,<sup>27</sup> a single subject experiment was used with with a 9-year old and a 14-year old boy.<sup>28</sup>

*Results*

The two boys attained clinically significant stuttering reductions at around 12-months follow-up beyond the clinic, with %SS scores of 3.0 or lower. Speech naturalness was measured with a 5-point scale where 1 = *very unnatural* and 5 = *very natural*. One boy’s mean scores at home changed from 1.8 pre-treatment to 4.1 at follow up, and the corresponding mean scores for the other boy were 2.8 and 4.1. For both boys, speech rate at home increased. These results suggest that treatment effects were not attained at the cost of unnatural sounding speech.

*A Phase II trial*

There were four adolescents among the mostly adult participants in the Phase II trial of self-imposed time-out overviewed during the previous lecture.<sup>29</sup> One withdrew because of no response to the treatment and another because the self-imposed time-out procedure could not be learned.

*Results*

The stuttering reductions of the remaining two adolescents were clinically significant. A 17 year old boy decreased stuttering from 3.2 %SS at pre-treatment to 1.1 %SS one week after the conclusion of instatement and generalisation. A 14 year old boy showed equivalent scores of 4.5 %SS and 1.0 %SS. Speech naturalness data were not presented specifically for these two adolescents, however all participants in the trial were assessed with the NAT scale, with the result that “around half of the participants achieved post-treatment speech naturalness scores within or near the range of control subjects” (p. 38).<sup>29</sup>

**Clinician-imposed time-out***Method*

There has been one clinical trial of a clinician-imposed, rather than self-imposed, time-out verbal response contingent model for school-age children.<sup>26</sup> Four children were recruited and three dropped out. There were the four treatment phases as described previously for GILCU.



*Results*

The mean pre-treatment stuttering of the four children was 7.6 stuttered words per minute. The child who remained in the trial had pre-treatment stuttering of 1.9 stuttered words per minute at follow-up. A total of 23.4 hours—much longer than GILCU—was required. The report contained no speech naturalness assessments, perhaps with the assumption that speech naturalness would not be an issue with a clinician-imposed time-out procedure.

**SYLLABLE-TIMED SPEECH****Method**

This treatment<sup>30</sup> is an adaptation of the Westmead Program for pre-schoolers discussed during Lectures Six and Seven. Two early reports suggested that the procedure may be useful with school-age children.<sup>31,32</sup> Another report, mentioned during Lecture Six,<sup>33</sup> exposed a group of 9–11 year olds to a metronome beat and their stuttering decreased without any instructions.

The treatment has two stages. During Stage 1 children use syllable-timed speech to establish a low level of stuttering, and the purpose of Stage 2 is to maintain those treatment benefits for a clinically significant period. During Stage 1 the children come to the clinic with a parent for a 30–45 minute session, during which they learn and practice syllable-timed speech. The child is taught to use the speech pattern to sound as natural as possible during these sessions. During the sessions parents are instructed to praise their children for using the speech pattern, and to remind them to use it if they lapse to a customary speech mode.

When the child is able to sustain the use of syllable-timed speech during a conversation, parents are instructed to practise the technique with the child for 5–10 minutes 4–6 times per day. When this is achieved parents are instructed to prompt their children to use the speech pattern at random times each day, at which time clinic visits occur fortnightly.

The treatment incorporates the SR scale, and the child is admitted to Stage 2 if %SS in the clinic is less than 1.5 and SRs for each day are 0–1 for two consecutive fortnightly clinic visits. During Stage 2 children are required to maintain those speech targets for 10–12 months with a performance contingent maintenance program.

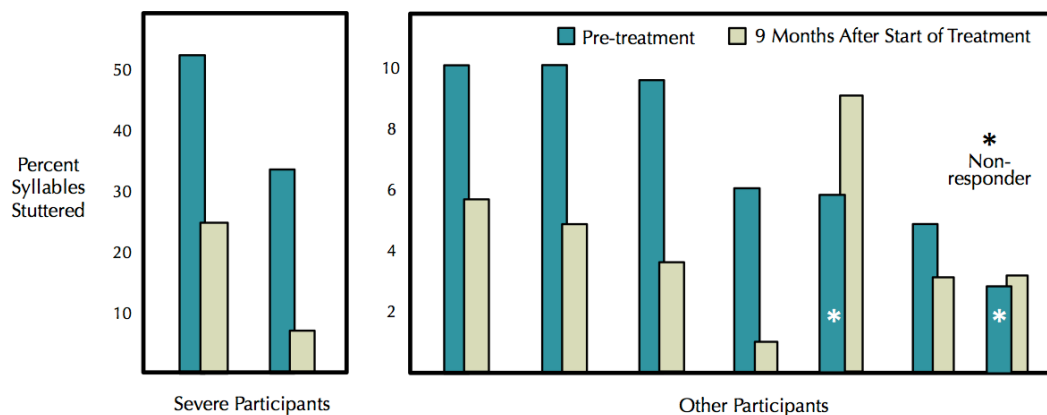
**A Phase I trial**

Ten children were recruited to a Phase I trial,<sup>30</sup> ages 6–11 years. One child dropped out from the treatment. Outcomes were measured at pre-treatment and at follow-up 9 months after the start of treatment.

**Results***Stuttering severity*

Two children did not respond to the treatment. Of the seven children who responded to the treatment, five reduced their stuttering by more than 50% at 9 months follow-up, and two children showed large reductions of 81% and 87% at 9 months follow-up. Mean reduction for the nine children was 54%. Results are shown in the figure.<sup>‡</sup> Two severe children are on the left and the remaining seven are on the right.

<sup>‡</sup> Adapted and reproduced with permission: Andrews, C et al (2012), Syllable-timed speech treatment for school-age children who stutter: A Phase I trial, *Language, Speech, and Hearing Services in Schools*, 43, 359–369, © 2012 American Speech-Language-Hearing Association.



### *Speech naturalness*

To assess speech naturalness, the experimenters obtained 15-second stutter-free speech segments from the children at follow-up, and played them to 10 unsophisticated listeners. The listeners were instructed to write down words or sentences to describe the children's speech. On only one occasion listeners gave any suggestion of a speech pattern change that might be associated with syllable-timed speech, with a report that one child sounded "mildly monotonal" (p. 365).<sup>30</sup> However, that child was one of those who did not improve.

### *Quality of life*

For the seven children who completed the treatment, mean speech satisfaction dropped from 6.0 pre-treatment to 2.7 at follow-up. Their quality of life scores on the OASES scale for this age group dropped from a mean of 54 to 40, showing a change from moderate to mild-moderate impairment.

## **A HYBRID TREATMENT: SYLLABLE-TIMED SPEECH AND VERBAL RESPONSE CONTINGENT STIMULATION**

### **Method**

The researchers who published the Phase I trial of syllable-timed speech for school age children<sup>30</sup> sought to improve the modest, and inconsistent, result of mean stuttering reduction around 50%. They proposed that a way to do that might be to add parent verbal contingencies to the treatment.<sup>34</sup>

The hybrid treatment is essentially the same as the syllable-timed speech treatment,<sup>30</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.

### **A Phase II trial**

Twenty-two children were recruited to the trial, ages 6–11 years, 16 of whom were boys and six were girls. Fourteen of the children had previous treatment, and nine had comorbid disorders, predominantly with speech and language. 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.<sup>‡</sup> The mean reduction of %SS scores at 12 months post Stage 1 completion was 77%. The

<sup>‡</sup> 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.

mean %SS score at 12 months post 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>30</sup> The group mean pre-treatment was 5.4 and 1.9 (SD=1.2) at 12 months post.

### Quality of life

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. That change represented an improvement from moderate to mild-moderate quality of life impairment.

## MACHINE AIDED TREATMENTS

### Electromyographic (EMG) biofeedback

#### Method

In short, 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 a number of tension-related disorders, and found its way into basic stuttering research with several promising laboratory reports.<sup>35,36,37,38</sup>

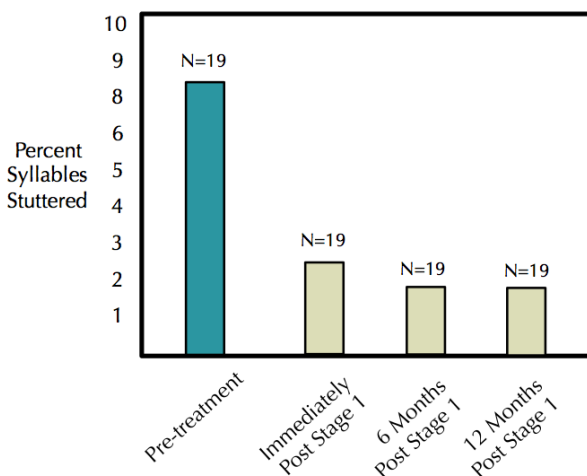
The trial of speech restructuring with 9–14 year olds described earlier, with and without parents present, contained an arm with EMG biofeedback:<sup>1</sup>

All children had their own computer so that speech muscle activity was monitored simultaneously ... if the child's muscle tension was too high a high-pitched sound would occur, indicating that the child should relax the speech muscles. Initially, the children were taught to raise and lower their muscle tension without speaking (around 2 hours). Once the child was able to distinguish differing levels of muscle tension ... he or she was required to perform the same task without observing the screen or being able to hear the sound.

Subsequently, the children followed a programmed instruction sequence lasting two days, starting with words and finishing with conversational speech. The programmed instruction included with and without EMG biofeedback during speaking. On the third day the children attempted speaking without the EMG biofeedback, and were able to enter a transfer phase when they could speak without stuttering and without biofeedback.

#### A Phase II trial

The arm with EMG biofeedback in that trial<sup>1</sup> involved five intensive 6.5-hour days. As with the other arms of the trial, it is not clear whether there were any drop-outs. Results in the figure,<sup>‡</sup> including follow-up,<sup>3</sup> show that the 5-day intensive treatment with the feedback produced results equivalent to those produced with intensive speech restructuring. At pre-treatment, mean stuttering severity was 11.5 %SS and at 12 months post-treatment was 2.9 %SS, showing a 75% stuttering reduction. The



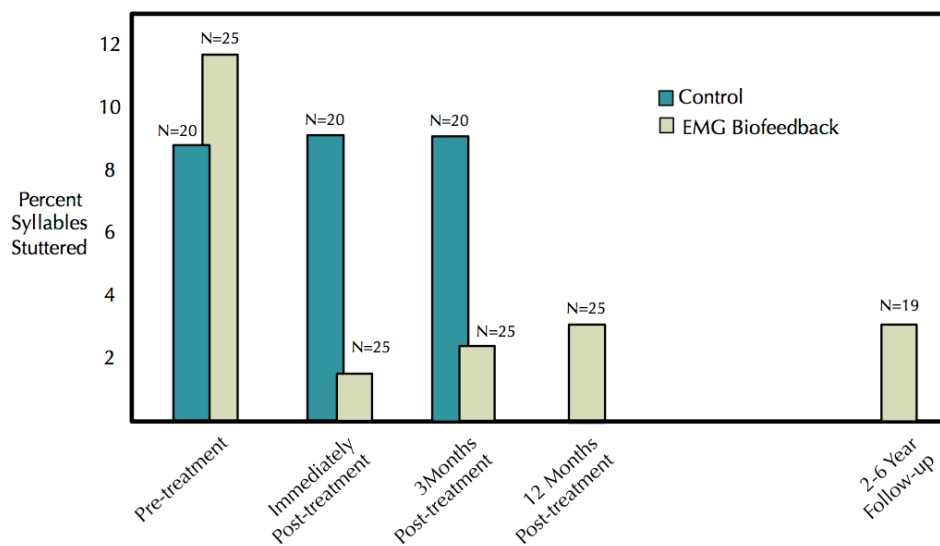
<sup>‡</sup> Adapted and reproduced with permission: Craig, A et al (1996), A controlled clinical trial for stuttering in persons aged 9 to 14 years, *Journal of Speech and Hearing Research*, 39, 808–826, © 1998 American Speech-Language-Hearing Association.

mean clinician pre-treatment speech naturalness score with the 5-point scale described previously was 2.6, and at post-treatment was 4.6, again suggesting that speech was reasonably natural sounding.

#### *Two failures to replicate*

The methods of the original trial<sup>1</sup> were replicated in another non-randomised trial<sup>39</sup> using identical equipment and following the biofeedback treatment manual.<sup>40</sup> However, results failed to replicate the original result. Participants were 10–16 years, which was a similar age range to the original study. Eleven of 12 participants recruited finished the study. Results showed only a modest mean stuttering reduction of 37% in conversations outside the clinic after the treatment.

Another non-randomised trial<sup>41</sup> again failed to replicate the positive findings of the original trial.<sup>1</sup> Three adolescents, one aged 13 years and two aged 15 years, were treated with a direct replication of the original study, again using identical equipment, and following the biofeedback treatment manual. Treatment effects were modest again, with %SS reductions for the three participants while talking on the telephone outside the clinic as follows, from pre-treatment to post-treatment: 12.8 to 9.1, 3.7 to 3.0, and 32.3 to 29.8.



#### **Altered Auditory Feedback**

There have been no clinical trials of the SpeechEasy device for adolescents or school-age children. However, within a Phase I trial<sup>26</sup> described earlier, four children were recruited to a speech restructuring procedure involving machine-based delayed auditory feedback. The trial was a failure, with all the children dropping out by the 9-month follow-up assessment.

### **CONCLUSIONS ABOUT THE ADOLESCENT AND SCHOOL-AGE EVIDENCE BASE**

#### **Speech restructuring**

For adolescents, speech restructuring has the strongest supportive clinical trial evidence of all available treatments, with independent replications. There is evidence that residential and non-residential treatment formats can be clinically useful. 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.

For school-age children, the evidence for speech restructuring is less compelling, with only one clinical trial reported<sup>1</sup> without independent replication, and three individual cases as described earlier. Another issue is that the clinical trial for school-age children contains participants 9 years and older. There is no clinical trial evidence that younger school-age children in the age range 6–8 years would benefit from the treatment. It is also a consideration that there is no clinical trial evidence of speech restructuring being viable for school-age children with a non-intensive format.

It is of interest, however, that a data-based case study<sup>42</sup> of a 1-week intensive speech restructuring treatment with fifteen 6–8 year olds showed clinically significant stuttering reductions in the clinic at 18-months post-treatment, and evidence of reasonably natural sounding speech. However, speech measures were not reported beyond the clinic. Another data-based case study without beyond-clinic data<sup>43</sup> contained six children ages 5–8 years. This was a 1-week intensive speech restructuring treatment, which used training with delayed auditory feedback. However, that report presented modest post-treatment stuttering reductions.

As with speech restructuring for adults, for adolescents and school-age children it is not possible without randomised controlled evidence to know anything about effect sizes that might be expected. 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 adolescent and school-age children.

There is evidence with adolescents that stuttering reductions with this style of treatment may or may not provide clinically significant anxiety reduction; at least with measures of situation avoidance. Consequently, adolescent clients may, or may not, require anxiety management in addition to stuttering reduction. Without sufficient participant numbers, however, it is currently not possible to know how often that would be necessary for adolescent clients.

### **Verbal response contingent stimulation**

For adolescents, there is almost no evidence that any of the verbal response contingent stimulation treatment models are suitable. The only clinical trial data involves two of four adolescent participants in an adult clinical trial of self-imposed time-out.<sup>29</sup>

For school-age children the outlook is a little more encouraging. A prominent finding is a positive non-randomised Phase II clinical trial of the Lidcombe Program model of verbal response contingent stimulation. One caveat is that there has been no independent replication of that finding. Another caveat is that the Lidcombe Program treatment model was developed for pre-school children, and so is unlikely to be completely suitable for older children.

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.

### **Syllable-timed speech**

There is no evidence for this treatment with adolescents. However, there is a Phase II non-randomised report for school-age children. On balance, it seems that the results of that trial are not as promising as for the Lidcombe Program. In contrast to the Lidcombe Program trial for school-age children,<sup>12</sup> the overall effect size appears to be more modest, and two children did not respond to the treatment at all.

On the positive side, this is by far the most procedurally simple of treatments for school-age children. Further, any treatment effects associated with syllable-timed speech seem not to be associated with unnatural sounding speech. Clearly, further clinical trials are needed to establish information for clinicians about the relative merits of the two treatments for school-age children. Those clinical trials will need to determine the extent to which stuttering reduction controls speech anxiety, because there is some suggestion that some school-age children will require anxiety management in addition to stuttering reduction.

**Machine-aided treatments**

After an encouraging clinical trial of EMG biofeedback for adolescents and school-age children, two subsequent clinical trials failed to replicate findings of any treatment effects. Considering that, and that EMG equipment and training to use it is required, this treatment cannot be recommended for adolescents or school-age children without further clinical trials.

**CLINICAL NOTES: 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 that period that span physical, cognitive, emotional and social domains. Those changes can impact the client and the family,<sup>44,45</sup> and can be a significant consideration when planning and implementing treatment.

Adolescents can be thought of clinically as neither children nor adults.<sup>46</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>47,48</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>44,49,50</sup> developing importance of the peer group<sup>47,51</sup> and a decline of motivation.<sup>52,53,54,55,56</sup> The emerging importance of the peer group is associated with a need to fit in to the norm,<sup>45</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>57</sup> is useful information for clinicians who treat this age group. An important finding was that some of them 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<sup>57</sup>

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)

However, the minority who reported unsupportive parent input found it to be “frustrating and perceived in an unsupportive manner” (p. 50).

**Telepractice and adolescents**

For adolescents, there are particular advantages to telepractice. Webcam treatment, which appears viable for adolescents,<sup>8</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 webcam treatment of their adolescent children. For example, they do not need to take them to and from the clinic. In particular, webcam telepractice 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>58</sup> Treatment methods for this age group can readily include technology<sup>59</sup> such as the Scenari-Aid website.<sup>60</sup>

## CLINICAL NOTES: SCHOOL-AGE CHILDREN

### A period of changing tractability

#### *The evidence for changing tractability*

Clinical trials of early intervention compared to clinical trials of adult intervention indicate that stuttering is at its most tractable shortly after onset, and at its least tractable during adulthood. Effect sizes are larger for pre-schoolers 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.

The Phase III clinical trial of the Lidcombe Program with pre-school children<sup>61</sup> shows no sign that %SS outcomes are affected by age. Table 5 (p. 662) of that 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 pre-school years.

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>13</sup> One of the children in that report, Participant 4, did not respond to the treatment, apparently because of compliance issues. For the remaining children in Table 2 (p. 284), ages 6–10 years, 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 those 11 children, 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 pre-schoolers 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.

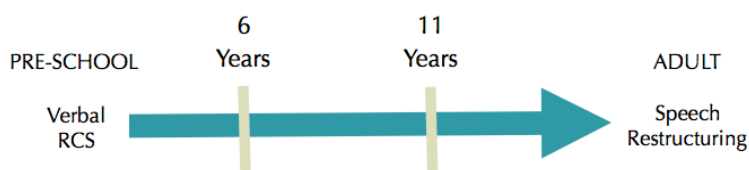
#### *A treatment selection model*

That reasoning 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.

Considering the clinical importance of the school-age time of life for stuttering, with its apparent changing clinical tractability, it is lamentable that there are no randomised clinical trials to provide guidance for clinicians. The gravity of the 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>62</sup> and more than 100 researchers and clinicians endorsed that call.<sup>63</sup>



<sup>†</sup> This analysis is not reported in the paper.

### **Adaptation of the Lidcombe Program for school-age children**

The model of treatment selection for school-age children in the diagram 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.

One group of Australian, American, and Canadians who use the treatment with school-age children comment as follows:<sup>64</sup>

Although school-age children's interests are often captured by the latest fad toys, we still find that more traditional games, toys, books, magazines, comics and catalogs continue to be useful as stimulus materials. (p. 153)

Those authors recommend that parent SR scores are used during the treatment, and that if the child contributes SRs they are used only as a supplementary source of useful information during treatment. They also recommend that during clinic visits there is open discussion with the child about the types, frequency and wording of verbal contingencies that will be used. They remark that school-age children generally prefer that verbal contingencies not be used in the presence of their peers. They also state that, because reading aloud in class is a routine part of school life, it is often useful to include reading aloud without stuttering as a therapy task. Additionally, they say, token rewards are more likely to be useful with this age group, although tick charts may be sufficiently motivating in many cases.

A common source of trouble with adapting the Lidcombe Program to this age group is the limited access parents have to children compared with pre-school age children. The authors of the article<sup>64</sup> suggest that in some cases when it is appropriate an older sibling, grandparent, or relative may be able to contribute to the treatment, providing that such a person attends all clinic visits.

### **Teachers and school-age children who stutter**

#### *The classroom*

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>65,66,67,68</sup> A common clinical picture of a school-age stuttering child is one who is quiet and withdrawn, and reluctant to participate in classroom activities, and is constantly anxious about being called on to speak in class. As expressed in an early report:<sup>69</sup>

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)

#### *The importance of teachers*

That early report<sup>69</sup> noted the importance of early school experiences because they often represent the first excursion into the world by children who stutter 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. Participants who were interviewed as young adults made it clear that the importance of the classroom experience extends into adolescence.<sup>70</sup>



*How might teachers help*

A useful teacher approach to fear of classroom speaking seems to have been originally suggested in 1940.<sup>71</sup> That paper contained sensible advice to teachers to not suggest 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>70</sup> indicated that such a constructive, individualised approach rarely occurred for the participants.

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, with this one being particularly informative:<sup>67</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)

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 from the back of the alphabet. Or, the teacher could call on children randomly, with the exception of 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>72</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 those 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>72</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>73</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>74</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>75</sup> Positive results were reported with a 9-year-old boy who included a classroom presentation about stuttering in his treatment,<sup>76</sup> and by speech-language pathologists who gave a classroom presentation as part of treatment for a 10-year-old girl.<sup>77</sup>

*Some modern resources*

Some modern suggestions about how teachers might help children who stutter in the classroom are available.<sup>78,79,80</sup> A video production by The Michael Palin Centre in London promotes teacher awareness of stuttering,<sup>81</sup> and is available at their website.<sup>78</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.

**SUMMARY**

Although there are no randomised controlled trials published for adolescent and school-age children who stutter, there are some clinically usefully non-randomised trials. There is reason to consider speech restructuring for adolescents and school-age children, supplemented with video self-modelling. For adolescents, webcam speech restructuring has been shown to be a viable and useful clinical method. For school-age children, there is reason to consider application of a suitably adapted version of the Lidcombe Program. There are some indications that self-imposed time-out may be useful with some adolescents and school-age children. For school-age children, there are indications that syllable-timed speech could be clinically useful. There is an encouraging nonrandomised trial showing that there are benefits from a hybrid treatment of syllable-timed speech and parent verbal contingencies. It seems clear that, for adolescents and school-age children, stuttering reduction is not necessarily associated with anxiety reduction as measured with situation avoidance. Clinical trial data suggest that verbal response contingent stimulation is progressively less suitable during the school years, with speech restructuring becoming progressively more suitable. Teachers are critical personnel in the lives of stuttering school-age children, and there are good reasons to include them in management plans.

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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>

###### *The current view*

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 those authors to conclude that there is compelling evidence of a relationship between stuttering and anxiety. The authors of that 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.

#### **ANXIETY**

##### **Expecting harm**

Generally speaking, expectancy of harm drives anxiety. Examples of exceptions to that generalisation include innate infant anxiety states such as fear of separation and 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 ignored. In fact, one report showed expectation of such social harm to be specifically the issue; those who stutter appeared to be troubled only by such expectations, not expectations of any physical harm.<sup>6</sup>

##### **Three components**

Anxiety is commonly described as a complex psychological event composed of verbal-cognitive, behavioural, and physiological components.<sup>1,7,8,9</sup> Clinical psychologists group the following cluster of emotions as related to anxiety: scared, shy, panicky, and insecure.

###### *Verbal-cognitive*

Anxiety commonly includes thoughts and expectancies about negative, harmful events. The prevailing psychological perspective is that, with the exception of 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 some kind of a negative view about them, and that negative view will cause them harm in some way, such as humiliating or demeaning them.

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<sup>†</sup> Thanks to Ross Menzies and Lisa Iverach for guidance with this material.

Naturally, most people will experience social anxiety in some situations, and 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 are “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 fleeing from 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 those three anxiety components—verbal-cognitive, behavioural, and physiological—as being loosely related to each other.<sup>8</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>10,11</sup> The latter report 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.

In particular, 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 or 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**

There is an extensive research literature, with much recent growth, showing that those who stutter are more socially anxious than controls.<sup>5</sup> Key findings from the last century were that those who stutter have high levels of anxiety according to standard clinical psychology measures,<sup>12,13</sup> and that stuttering and control subjects can be accurately distinguished using such measures without any speech data.<sup>14,15</sup> The former report<sup>14</sup> used the Speech Situation Checklist 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>14</sup> For 21 selected situations, the reported strength of such emotions was able to predict with 93% accuracy whether participants stuttered or not. That result was replicated in the later study<sup>15</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.

Research findings this century continue to confirm that, as a group, adults who stutter are socially anxious compared to those who do not.<sup>9,16,17,18,19,20</sup> A recent meta-analysis<sup>21</sup> confirmed this for trait anxiety and social anxiety. Studies were included in the analysis if they used proven psychometric measures of trait and social anxiety, and if they compared a stuttering group of participants to a control group. Eleven studies dealt with trait anxiety and eight dealt with social anxiety. Findings confirmed that those who stutter differ as a group from those who do not stutter, with an effect size (see Lecture Five) of  $d=0.57$  for state 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 possibly—but by no means certainly—will have a history of social anxiety.

Salivary cortisol is a physiological marker of anxiety which has been associated with the disorder several times.<sup>22,23</sup> The most recent study of 19 adult men who stuttered and 19 matched controls<sup>24</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 that result, a physiological measure of unstimulated saliva flow rate was lower for the stuttering participants than the controls.

## Stuttering and social anxiety disorder

### *What is social anxiety disorder?*

Social anxiety disorder, also known as social phobia, is described in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5)<sup>25</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>26,27,28,29</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>30</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 the expectation of humiliation and embarrassment in social situations is unrealistic and irrational in light of the actual threat.

### *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 the disorder is 8–13%.<sup>31,32</sup> However, case reports of social anxiety disorder are common for those who stutter,<sup>33,34</sup> with the condition reported for 40%,<sup>35</sup> 44%<sup>36</sup> and 60%<sup>37</sup> of cases in speech clinics. The latter report was that such cases have 34-fold increased odds of meeting criteria for social anxiety disorder diagnosis compared to age and gender matched community controls. Those reports are consistent with studies reporting that adults who stutter in general have anxiety scores higher than controls but slightly lower than those with psychiatric conditions.<sup>13,19,38</sup> However, a report of older stuttering participants after a lifetime with the disorder<sup>39</sup> did show anxiety scores in the range associated with social anxiety disorder. A recent review of social anxiety disorder and stuttering is available.<sup>31</sup> A report compared the demographics of clients presenting to speech clinics for stuttering treatment with and without social anxiety disorder.<sup>40</sup> Apart

from the group with social anxiety disorder being significantly younger, no demographic differences were found.

#### *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 reaction at some stage because of their stuttering. 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. 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>41</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. 16). The list included “people have been unkind to me,” “people have avoided looking at me,” and “people made fun of me or picked on me” (p. 36). Yet, the majority of 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. 38).

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>42</sup> showed that many store clerks, when spoken to by someone stuttering, reported that they experienced embarrassment, pity, and sympathy. There were some early reports of physiological listener responses to stuttering, and a more recent one<sup>43</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>44,45,46</sup> that show listeners looking away from videos of people stuttering more often than for control speech. A report<sup>47</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 to 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**

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 those also.

There are reports<sup>48,49</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 anxiety related: obsessive compulsive disorder, dependent personality disorder, and avoidant personality disorder. However, another report about stuttering and personality disorders,<sup>50</sup> which used a different assessment than the report just mentioned,<sup>48</sup> failed to find any evidence of personality disorders among adults seeking treatment for stuttering.

A study of 200 stuttering and control adults<sup>20</sup> with the Symptom Checklist–Revised (SCL-90-R)<sup>51</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>52</sup> reported the relation 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.

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>49</sup> That finding was replicated with a community

sample within a British birth cohort study<sup>53</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.

## 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 to them. 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

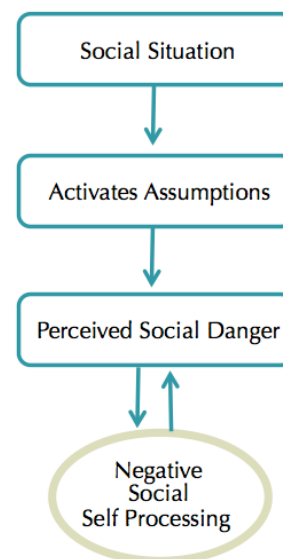
A major contribution of this model<sup>26</sup> is that it explains that puzzle about social anxiety disorder; why social anxiety persists in the face of constant experiences that should disconfirm the belief that social situations are harmful. There are many models to explain this,<sup>54,55,56,57,58,59</sup> and one has been devised specifically for stuttering.<sup>60</sup> However, the Clark and Wells model is the most influential, having been confirmed several times with tests of hypotheses derived from it. Several cognitive models of social anxiety have been developed and validated, but the Clark and Wells model dominates 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>61,62,63,64</sup> Components of the model feature in a treatment for the social anxiety of those who stutter discussed during the next lecture. The model has been overviewed in an easily understandable way,<sup>65</sup> and that overview is essential reading for speech-language pathologists who commonly encounter stuttering clients with social anxiety. The following description of the model draws heavily from that source.

### Three assumptions of the model

The model rests on three assumptions about those affected. The first is 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>65</sup> The second assumption is 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>65</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>65</sup> Such assumptions cause a perception of an impending social situation to be threatening.

### Negative self processing in social situations

An impending social situation activates the assumptions just described, as shown in the figure,<sup>‡</sup> which signal that the impending social situation is threatening, making it a source of perceived social danger. Then, during the social encounter, negative self-processing prevents disconfirmation of the



<sup>‡</sup> 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 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 how people think what 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>66,67,68,69</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 that situation, because it is impossible to see an observer perspective of yourself, only 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>65</sup>

Findings about observer perspective have been reported with interviews of adult stuttering and control participants.<sup>70</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>71</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 the 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:<sup>71</sup>

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)

### *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>72</sup> detect negative social information more accurately than positive social information,<sup>73</sup> have slow recognition of positive social stimuli,<sup>74</sup> and pay excessive attention to emotional social stimuli.<sup>75,76</sup>

There has been a finding to that effect with stuttering participants,<sup>77</sup> who looked less often than controls at positive faces while speaking to a group. They also looked less often and for a shorter time to all audience members compared to controls. The results of Stroop task<sup>†</sup> studies<sup>78,79</sup> are broadly consistent with that finding. The latter study reported that, compared to controls, participants in the stuttering group showed bias toward socially threatening words such as “inept,” “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. However, one report has failed to show an effect for stuttering participants; at least for those who were not socially anxious.<sup>80</sup>

<sup>†</sup> Stroop tasks assess reaction time interference. Participants name the colours of the text in which different words are printed.

So, if someone is socially anxious and perceives social situations as dangerous, a destructive cycle of negative processing of thoughts can begin during the social encounter and over subsequent encounters that makes the perceived threat worse. Overall, this intensifies the negative self-processing. That destructive cycle is shown with the bidirectional arrows in the figure.

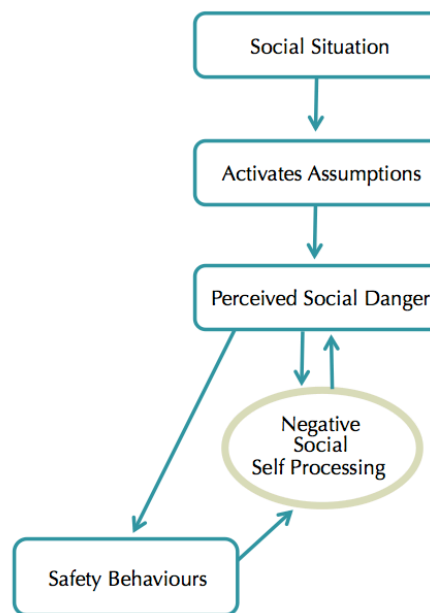
## Safety behaviours

### *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 is presented in the Subtle Avoidance Frequency Questionnaire.<sup>81</sup>

### *Safety behaviour prevent fear extinction*

There is evidence<sup>82,83,84,85,86</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 doing so 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 social danger can feed into a cycle of worsening perceived social danger, as shown in the figure.<sup>‡</sup>

### *Eliminating safety behaviours during anxiety treatment*

Safety behaviours are routinely targeted for elimination during anxiety treatment.<sup>26,65,87</sup> A review<sup>88</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>89</sup> There is evidence that this occurs with speech-language pathologists who treat stuttering clients.<sup>90</sup> Based on a survey of 160 speech-language pathologists, there is evidence that they may recommend what might, in effect, be safety behaviours to adult clients in order to

<sup>‡</sup> 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.

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.

The most commonly recommended of the *General Safety Behaviours* was avoiding anxiety provoking topics. Clinical psychologists recognise that strategy as a common safety behaviour intended to protect against social threat. Clinical psychologists also recognise silent rehearsal before speaking as safety behaviour, and the majority 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.

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 in the event that clients do use them. In other words, there is a need to determine whether they are adaptive and helpful as described below, or in fact prevent fear extinction.

In the interim until such research is done, the results of that study<sup>90</sup> need to be kept in mind by clinicians during use of speech restructuring treatments with clients. It is possible to place clients in a situation where they attain control of stuttering at the expense of perpetuating speech related anxiety. That being said, in clinical psychology it is not currently clear what client behaviours should be considered safety behaviours and what behaviours should be considered healthy adaptive behaviours in a situation that causes anxiety.<sup>82,89</sup> That currently is the situation with stuttering treatment.

<i>SAFETY BEHAVIOUR CATEGORY</i>	<i>EXAMPLES</i>
<i>General Safety Behaviours</i>	<p><i>Avoid topics that make you anxious</i></p> <p><i>Ask many questions</i></p> <p><i>Point rather than speaking</i></p> <p><i>Allow your partner to talk for you</i></p> <p><i>Talk little</i></p>
<i>Practice and Rehearsal</i>	<p><i>Rehearse mentally before speaking</i></p> <p><i>Practice the speech restructuring technique just prior to speaking</i></p> <p><i>Rehearse answering the phone mentally before answering</i></p>
<i>General Avoidance</i>	<p><i>Avoid unnecessary talking on a bad day</i></p> <p><i>Keep answers short</i></p> <p><i>If anxious avoid difficult words</i></p>
<i>Choose Safe and Easy people</i>	<p><i>Immediately before an important speaking situation</i></p> <p><i>In socially threatening situations</i></p>
<i>Control Related</i>	<p><i>Say "relax" to yourself when anxious</i></p> <p><i>Speak slowly when anxious</i></p> <p><i>Try to take deep breaths</i></p>

A subsequent report<sup>91</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 commonly 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 reported use of many of the safety behaviours was 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.

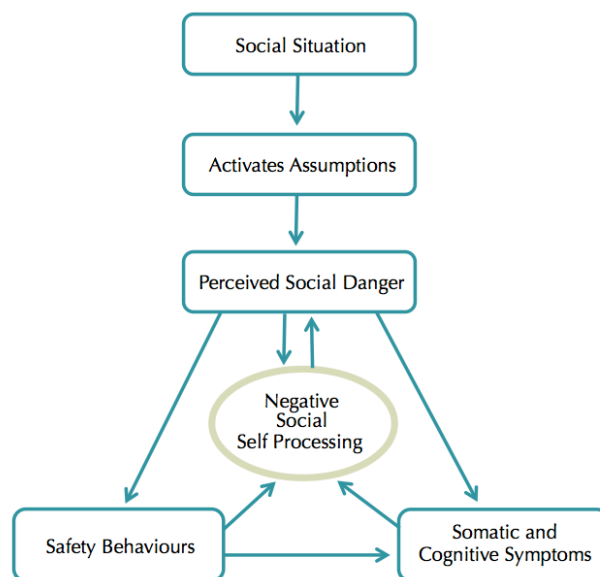
In this context, it is worth considering the commonly recommended technique of self-disclosure with clients who stutter. This is where, at the start of a social interaction or address to unfamiliar listeners, the speaker announces that stuttering is likely to occur. Common reasoning for the use of this strategy is the likelihood of listener stereotyping of stuttering, as described during Lecture Two. In which case, if it is done to prevent such listener response, it is a safety behaviour. There is evidence that self-disclosure so can positively influence listener reaction,<sup>92,93,94,95,96</sup> so there is a need to determine whether the technique in fact prevents fear extinction or has the effect of exacerbating the feared outcome.

### 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:<sup>‡</sup> sweating, blushing, increased heart rate, heart palpitations, hyperventilation, shortness of breath, nausea, headache, shaking, feeling flushed, and muscular tension. Those 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 was not at all based in 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.



<sup>‡</sup> 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.

### After the feared situation

After the event those with social anxiety disorder can conduct a “post mortem of the event” (p. 411).<sup>65</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:<sup>65</sup>

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)

## ANXIETY IMPAIRS SPEECH TREATMENT

### The problem of post-treatment relapse

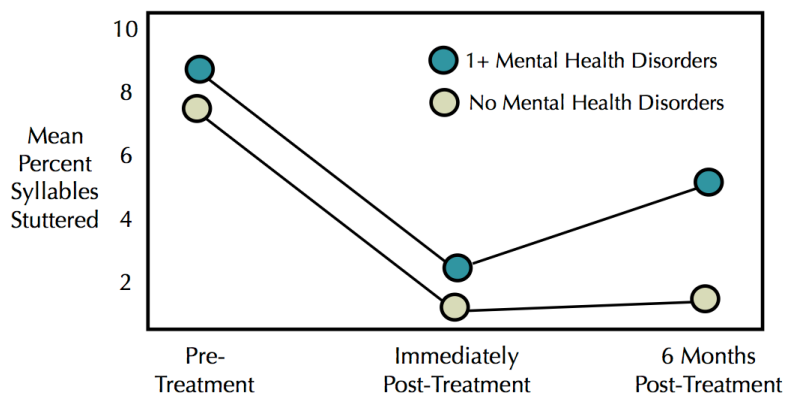
For decades it has been known that after speech restructuring treatment to reduce or eliminate stuttering only around one third of clients are able to sustain their treatment benefits.<sup>97,98</sup> In other words, the relapse rate after speech treatment is around two-thirds:<sup>97</sup>

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)

### Anxiety and post-treatment relapse

No plausible explanation for that relapse rate was forthcoming until a publication linked it to anxiety, showing that the one-third of clients with self-reported relapse had elevated anxiety scores on the Spielberger Trait Anxiety Scale.<sup>98</sup> This was verified with a study of 64 adults who received a speech restructuring treatment in clinical trials.<sup>99</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.<sup>‡</sup> 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, these clients had more severe stuttering than those who had no mental health disorders. They also were a less clinically responsive group. They reduced 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



<sup>‡</sup> 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.



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 ORIGINS OF SOCIAL ANXIETY WITH STUTTERING: THE PRE-SCHOOL YEARS**

Obviously, the frank social anxiety problems that trouble some adults who stutter begin some time earlier in life. It is important, then, when pre-school children present to clinics with stuttering, to identify any signs that might signal the potential for future development of social anxiety problems.

### **Direct evidence: Psychometrics**

One report<sup>100</sup> incorporated data for eight children who showed pre-treatment scores in the normal range for the Child Behaviour Checklist,<sup>101</sup> which is a parent report measure that includes behavioural markers of anxiety. Another report<sup>102</sup> presented data for seven stuttering and seven control children and showed that they did not differ on the Preschool Anxiety Scale,<sup>103</sup> which is a more direct parent report measure of anxiety. The report also showed no differences for salivary cortisol. However, those two reports do not contain enough participants to be particularly convincing.

The Pictorial Scale of Perceived Competence and Social Acceptance for Young Children<sup>104</sup> is a direct assessment based on child responses. It was given to 28 children who stuttered, mean age 4 years 9 months, and a control group of children.<sup>105</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.

The report for the ELVS cohort (see Lecture Two) at 4 years of age<sup>106</sup> presented data from the Pediatric Quality of Life Inventory Parent-Proxy Report (PedsQL),<sup>107</sup> which is a medically oriented quality of life scale completed by parents. It showed, at 4 years of age, that no differences for “psychosocial health related quality of life” (p. 464)<sup>106</sup> were associated with stuttering. Again, there are qualifications about interpretation of those data because the PedsQL scale is not a standard measure of childhood anxiety used in the 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>107</sup>

Another report<sup>108</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 (see Lecture Seven). Compared to normative data, there were no systematic problems for the children according to four medically oriented quality of life instruments.

The ELVS report<sup>106</sup> also featured the Strengths and Difficulties Questionnaire, which is an assessment used commonly in child mental health research.<sup>109,110,111</sup> It measures emotional and behavioural problems with five scales: “emotional symptoms, conduct problems, hyperactivity-inattention, peer problems, and prosocial behavior” (p. 1337).<sup>110</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. However, another report<sup>112</sup> provided contrasting results for the Strengths and Difficulties Questionnaire using the Millenium Cohort.<sup>113</sup> The Millenium 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 their children stuttered. At those ages parents were also asked to complete the Strengths and Difficulties Questionnaire, along with many other assessments. Data were available for 173 three-year-olds, 194 five-year-olds, and 170 eleven-year-olds reported to be stuttering. The Total Difficulties scores showed statistically and clinically

significant differences 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).

The Millenium Cohort publication acknowledges the limitations of the parent report method for identifying stuttering compared to the direct diagnostic method of the ELVS cohort. Parent reports of stuttering in a questionnaire are not particularly compelling, and obviously underestimated prevalence (see Table 1, p. 27). That being said, significant findings between stuttering and nonstuttering children for the Strengths and Difficulties Questionnaire so early during the developmental course of stuttering needs to be considered when forming a clinical view about when to begin early intervention for the disorder, as discussed shortly.

### **Direct evidence: 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, constitutional tendency to react to the environment or interact with it in a certain fashion. It is caused by biological features that include genetics. Childhood temperament is generally accepted as a risk factor for anxiety later in life.

The most commonly used classification of childhood temperament was developed by Thomas and Chess,<sup>114</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 stuttering: Some fundamental caveats*

There is a rapidly accumulating body of literature, which is turning out to be controversial, dealing with the association between early childhood temperament and stuttering. In order to properly interpret that literature, it is essential to understand its inherent limitations.

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. As outlined during this lecture, it is clear that pre-school children may encounter all kinds of adverse psychological events after stuttering onset. Therefore, it is 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 the likelihood that, in part, they reflect experiences of stuttering. In short, the essence of this caveat is that any association between early stuttering and measures of temperament may reflect the effects of stuttering rather than its causes.<sup>119,115</sup>

A second caveat with this literature is that it is observational, not experimental. In other words, it is based on 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 definitely establish anything about the causality of stuttering; it can only suggest the possibility a causal connection. A third caveat is that any findings of association between temperament and early stuttering may pertain to speech and language disorders generally rather than to stuttering specifically.<sup>116,117,118</sup> And finally, measures of temperament for pre-schoolers are based on parent report, which may be influenced by the presence of stuttering.

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

### Three reviews of early childhood temperament and stuttering

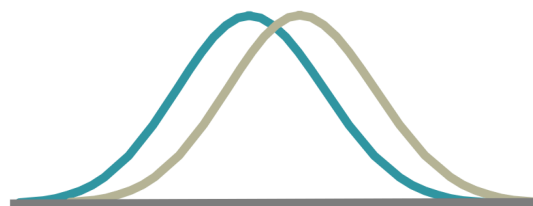
A full review of the topic is beyond the scope of this lecture, but the pertinent research is cited in three current reviews.<sup>118,119,120</sup> One of them<sup>118</sup> cites evidence that a generally so-called “difficult” temperament heightens the risk of anxiety disorders later in life. “Difficult” temperament includes “nervous, high strung or tense,” “appears fearful or anxious,” “appears worried,” “not as happy as other children,” “has difficulty having fun” (p. 153).<sup>118</sup> The review concluded:<sup>118</sup>

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)

Another review<sup>119</sup> was published 2 years later, incorporating 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>119</sup> In another publication,<sup>121</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 to controls, they stated that children who stutter are (1) less adaptable, (2) have poorer attention and attention regulation, and (3) have a negative mood.

The authors of the review<sup>119</sup> go as far to suggest, with considerable caution, that there may be clinical implications of such an association. 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 present preliminary results suggesting that temperament predicts outcome of an “indirect” therapy.<sup>122</sup>

The most recent review<sup>120</sup> of the topic focused on the effect sizes reported in the literature, and recorded 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 around  $d=0.60$ . The review author makes the point that the two distributions for such an effect size are generally not apparent to readers, but look something like the diagram. There is extreme overlap between the two groups and no real separation between them. All this led the reviewer to conclude:<sup>120</sup>



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)

### Parent measures of temperament

The prospective ELVS cohort was used to determine the presence of temperament anxiety markers prior to and after the development of stuttering.<sup>123</sup> That report involved 183 children who stuttered and 1,261 nonstuttering children 2–4 years of age. At the children’s second, third and fourth birthdays, parents completed the Short Temperament Scale,<sup>124,125</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 children who stuttered “were less reactive to environmental stimuli and had a reduced ability to attend to a task until completion” (p. 1314).<sup>123</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<sup>123</sup>

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)

The second Illinois cohort<sup>126</sup> (see Lecture Two) studied 58 stuttering pre-school children and 40 control children. According to one subtest of the Children's Behavior Questionnaire-Short Form,<sup>127</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 relation between early stuttering and temperament, there could be a correlation between stuttering severity and temperament measures. Nine studies have reported such a correlation or association<sup>128,129,130,131,132,133,134,135,136</sup> and three have failed to show a correlation.<sup>137,138,139</sup> A recent study along those lines<sup>134</sup> involved 69 children who stuttered, mean age 3 years 7 months. The authors sought to correlate the temperament dimension of "effortful control" with parent measures of stuttering severity. The effortful control temperament dimension describes capacity to regulate focus and attention, and was measured with the Children's Behavior Questionnaire-Short Form.<sup>140</sup> A regression model found a statistically significant relation between stuttering severity and effortful control, with 20% of the variance explained. Another recent report<sup>135</sup> with 47 pre-schoolers 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. However, only around 10% of the variance was explained. That report failed to find any similar correlations for skin conductance.

The authors of one of the recent reports<sup>134</sup> expressed a view consistent with the authors of the autonomic nervous system study reviewed earlier,<sup>121</sup> that such an effect would contribute to sustaining early stuttering by influencing stuttering severity. They suggested the following, perhaps controversially:<sup>134</sup>

Developing treatment approaches from the platform of temperament appears to be a promising strategy. This would emphasize that treatment for stuttering in children should include skills related to the self regulation of EC [effortful control] (i.e., attention, inhibitory control, and perceptual sensitivity). (p. 91)

#### *Autonomic nervous system studies*

Two studies have been reported comparing the autonomic nervous systems of pre-schoolers who stuttered and controls.<sup>121,141</sup> The first report studied 20 pre-schoolers who stuttered 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.

The children watched a neutral screen for a while during a baseline condition. 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 children who stuttered showed lower (parasympathetic) respiratory sinus arrhythmia during the baseline, which theoretically means they had increased vulnerability to a sympathetic response. Additionally, the children who stuttered showed more (sympathetic) skin conductance increase during positive emotions while watching and talking about videos. Interestingly, a maximum effect size of  $d=0.62$  was reported (Table 3), consistent with the observations in a previous review.<sup>120</sup>

<sup>†</sup> The sympathetic nervous system controls responses to perceived threat, and the parasympathetic nervous system controls homeostasis at rest.

The authors concluded that their results suggested autonomic nervous system involvement with early 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 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 that 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 stuttering,<sup>141</sup> those authors again presented that 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 pre-school children with persisting stuttering and 23 who they considered recovered around 2 years later.<sup>142</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 that “that emotion should be considered in any comprehensive account of childhood stuttering,” regardless of “the directionality of effect” (p. 149).

#### *Executive functioning: Response inhibition and attentional control*

The term executive functioning refers broadly to cognitive activity that exerts control over day-to-day functions, including thoughts, emotions and behaviour. One component of executive functioning is response inhibition, also known as inhibitory control. 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. With pre-school children, the day-night task is commonly used, where children are asked to say “day” when shown a picture of night time, and asked to say “night” when shown a picture of day time.

A nonverbal version of the day-night task—a grass-snow task—requires children to point to a white card when they hear the word “grass” and to point to a green card when they hear the word “snow.” That nonverbal grass-snow task inhibition paradigm was used in a study of 41 children who stuttered and 41 controls aged 3–6 years.<sup>143</sup> Additionally, a version of the paradigm was used—a baa-meow task—that involved no verbal stimuli. The children were played sheep and cat noises and asked to push the button that was the opposite of the animal they heard.

Results showed that the stuttering group was significantly less accurate than controls for the baa-meow task, but not for the grass-snow task that involved a verbal stimulus. For both tasks, the children in the stuttering group had slower reaction times. Significant correlations were reported between the accuracy and reaction time scores and parent scores on the Inhibitory Control scale of the Children’s Behavior Questionnaire-Short Form.<sup>127</sup> The authors concluded that the results “suggest that response inhibition is an important aspect in describing the underlying nature of stuttering in early childhood” (p. 848). Naturally, results could have been a result of stuttering, and the authors did not intend an implication that difficulties with inhibition relate to the causes of stuttering, but they certainly would not dismiss that possibility.<sup>144</sup>

Response inhibition (also known as inhibitory control) was studied for a group of 16 Finnish-speaking children who stuttered and 16 controls, with mean ages 7.1 years.<sup>145</sup> A button-press reaction time task was used with three response variations: (1) a standard response of hearing a single, low-pitched tone and pushing the button once, and hearing a double low-pitched tone and pushing the button twice, (2) an “incompatible” (p. 3161) response of hearing a single, high-pitched tone once and pushing the button twice, and hearing a double, high-pitched tone and pushing the button once, and (3) a “mixed” (p. 3161) response combining the two formats. Some evidence was found for reduced response inhibition for the stuttering group.

That report<sup>145</sup> also found a result for attentional control, which is also known as attentional shifting; broadly speaking being the capacity to concentrate on one thing. In terms of errors made with responding, there was some evidence for impaired intentional control for the stuttering group. In

relation to the issue of whether results pertained to the causes or the effects of stuttering, the authors state that their report and others about related topics “have contributed to a better understanding of their possible causal or exacerbating role in the development of stuttering” (p. 3167).<sup>†</sup>

Another research group<sup>146</sup> studied executive functioning with a group of 75 stuttering pre-schoolers and 75 controls. The children were assessed with a parent scale, the Behavioral Rating Inventory of Executive Function-Preschool Version,<sup>147</sup> and a behavioural task, the Head-Toes-Knees-Shoulders. The latter task requires children to do the opposite of the examiner instruction. For example, when asked to touch their head they are required to touch their toes. Data suggested poorer executive functioning scores for the stuttering children, even to the extent of being more likely to show clinically significant differences. Also, the stuttering children performed more poorly than controls on the behavioural task. Perhaps controversially, the authors concluded

... although we must proceed with caution when applying these findings to everyday clinical practice, clinicians could consider introducing standardized parent-report scales of EF [executive functioning] as part of the fluency diagnostic for young CWS [children who stutter] ... Such an assessment may reveal concomitant EF difficulties in some CWS that may need to be considered when developing a comprehensive, holistic treatment plan. (p. 29)

### Indirect evidence

#### *Parent-reported child awareness and negative peer reactions to early stuttering*

The potential anxiety issues reported for adults who stutter can be attributed to negative social conditioning during peer interactions early in life.<sup>26,28</sup> Consequently, it is important to recognise a body of research suggesting that pre-school children are likely to be aware of their stuttering and that it is capable of causing them distress. That 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 experimenter during a 6-year period documented this effect.<sup>148</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.<sup>148</sup>

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)

In another early report,<sup>149</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,<sup>150</sup> and the figure had increased to 90% for 7-year olds. Around two-thirds of children who had been stuttering for less than 1 month reportedly showed awareness of it. A report of 77 parents of children, mean age 53 months (range 34–73 months),<sup>151</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,

<sup>†</sup> There is an intriguing caveat to the findings of this study, and any other that involves responses of rapid finger-press sequences by those who stutter. There is evidence of impaired finger movement sequences with stuttering (see Lecture Three). Hence that, as well as anomalies of executive function, might explain subtle differences between the groups.

withdrawal, reduced or changed verbal output, making comments about their inability to talk" (p. 407).<sup>151</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 than the usual 6–22% range reported for children who do not stutter.<sup>152,153</sup>

### *The KiddyCat*

The KiddyCat<sup>154</sup> is a scale developed from a version for older children, to be discussed shortly. Its development<sup>155</sup> was driven by the Anticipatory Struggle Hypothesis,<sup>156</sup> which is a theory that is currently outdated, mentioned during Lecture Three. The Anticipatory Struggle Hypothesis posits that stuttering is driven and sustained by an early developing belief that speech is difficult. The KiddyCat 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," and "do people like how you talk?" (p. 229).<sup>157</sup> Consequently, it might be interpreted as having some relation to social anxiety.

Three studies have shown KiddyCat score differences between stuttering and control pre-school children in the United States: with 52 stuttering and 62 control children,<sup>157</sup> 45 stuttering and 63 control children,<sup>158</sup> and with 46 stuttering and 66 control children.<sup>159</sup> The same result has been reported for 58 stuttering and 70 control Polish children,<sup>160</sup> and for 49 stuttering and 74 control Slovenian children.<sup>161</sup> The KiddyCat was used in a study mentioned previously with small participant numbers,<sup>102</sup> and showed no differences between stuttering and nonstuttering children.

### *Evidence of negative peer responses*

There is direct video evidence of negative peer reactions to stuttering in pre-school playgrounds.<sup>162</sup> Four pre-school children were video recorded during four 20-minute playground periods. During those 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 pre-school 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 stuttering pre-school children may receive negative social conditioning comes from an ingenious research paradigm involving stuttering and nonstuttering puppets. 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" (p. 233–234).<sup>163</sup> 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>164</sup> by adapting 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 Two. Preliminary results with 51 children 3–7 years old indicate the potential for negative attitudes toward the disorder at that time of life.

## **Conclusions**

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 stuttering may be associated with negative peer social conditioning that 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, with the ELVS cohort showing normal scores for the Strengths and Difficulties Questionnaire during the pre-school years, but the Millenium cohort showing abnormal scores. One prospective study, although yet to be replicated, 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. It may be that the social anxiety that troubles many who present to clinics with chronic stuttering is not at all driven by innate genetic factors, but only by social conditioning that occurs after stuttering onset.

## **THE ORIGINS OF SOCIAL ANXIETY WITH STUTTERING: THE SCHOOL-AGE YEARS AND ADOLESCENCE**

### **Direct evidence**

#### *General Anxiety Scale for Children*

The 1,000-family study<sup>165</sup> (see Lecture Two) presented extensive early data about anxiety with a group of 80 school-age children who stuttered 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>166</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>167</sup> In the context of a clinical trial, the State-Trait Anxiety Inventory for Children<sup>168</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>169</sup> replicated that 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>170</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 are used in the test.

Another report<sup>171</sup> used the State Trait Anxiety Inventory<sup>172</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>171</sup> used the long version of the Fear of Negative Evaluation scale<sup>173</sup> with adolescents (mean age 14.4 years), and reported significantly higher scores for the stuttering group. However, a caveat to that finding is that the Fear of Negative Evaluation scale was developed for adults, not adolescents.

#### *Revised Children's Manifest Anxiety Scale*

Revised Children's Manifest Anxiety Scale<sup>174</sup> data were reported with 18 stuttering and 18 control children in the age range 11–12 years.<sup>175</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 children who stuttered were not beyond the normal range of scores. Another report<sup>176</sup> with 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 normal range for anxiety.



A report<sup>177</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 normal 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>178</sup> deals with fear of speaking, with subscales for public speaking, meetings, groups and conversations. A study<sup>179</sup> recruited 39 stuttering adolescents from speech clinics along with 39 controls, ages 13–18 years with a mean of 14.6 years. The stuttering adolescents showed significantly higher apprehension scores than controls. This result was replicated with a study<sup>180</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>179</sup>

#### *Strengths and Difficulties Questionnaire and Coping Inventory for Stressful Situations-Adolescent*

The Strengths and Difficulties Questionnaire<sup>110</sup> is a general assessment of psychological health, and the Coping Inventory for Stressful Situations-Adolescent measures responses to stressful situations. A report with 35 stuttering boys and 35 controls, ages 14–17,<sup>181</sup> found no overall differences between the groups, although the stuttering boys scored in the high end of the normal range. Significant results were reported for some subtests. For both tests the stuttering boys had significantly more peer relationship problems, and for the latter test they had significantly fewer socially desirable behaviours.

#### *Multiple Anxiety Assessments*

A report<sup>182</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>183</sup> Fourteen of them (38%) met diagnostic criteria for at least one DSM-IV<sup>184</sup> mental health disorder,<sup>†</sup> which is around twice the anticipated rate for adolescents, which is in the 17–21% range.<sup>185,186,187</sup> Ten of those 14 diagnoses were a mental health disorder involving anxiety: social anxiety disorder, agoraphobia, obsessive-compulsive disorder, separation anxiety disorder, and specific phobia.

For four psychological test scores the stuttering participant scores were in the normal range: Revised Children's Manifest Anxiety Scale,<sup>174</sup> Youth Self Report and Child Behavior Checklist<sup>188</sup> and the Children's Depression Inventory.<sup>189</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 study of 75 stuttering 7–12 year-olds and 150 matched nonstuttering controls<sup>190</sup> included the Youth Online Diagnostic Assessment.<sup>191</sup> Results indicated that, compared to controls, the stuttering group had four-fold increased odds for prevalence of any anxiety disorder. For social anxiety disorder there was a six-fold increased odds, with girls much more at risk than boys, with 24% of the children who stuttered diagnosed with the condition.

The Spence Children's Anxiety Scale Child Report<sup>192</sup> and Parent Report<sup>193</sup> showed scores within the normal range but significantly higher values for the stuttering group. Mean scores for the Strengths and Difficulties Questionnaire Parent Report<sup>194</sup> were within normal limits, however the mean Total Difficulties score and Internalising and Externalising scores were significantly higher for the children who stuttered.

The largest cohort to date<sup>195</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>174</sup> the Children's Depression Inventory,<sup>189</sup> the Youth Self Report and Child Behavior Checklist,<sup>188</sup>

<sup>†</sup> The DSM-IV is the previous edition of the DSM-5, which was mentioned previously.

and the Assessment of the Child's Experience of Stuttering, which is an earlier version of the OASES-S (see Lecture Four). 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 normal 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>196</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>192,193</sup> the Strengths and Difficulties Questionnaire,<sup>110</sup> and the School-Aged Temperament Inventory.<sup>197</sup> In contrast to the study of other Australian children reported around the same time,<sup>195</sup> no differences were found. A likely cause of that discrepancy is that one study involved clinical children and the ELVS report did not.

#### *Physiological evidence*

A study<sup>198</sup> of nine children who stuttered, 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 abnormal levels were 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 Two). 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 Two). 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>199,200,201</sup>

### **Indirect evidence**

#### *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>202</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. For the same reasons that the KiddyCAT is of interest in the context of potential anxiety development for pre-schoolers, the Communication Attitude Test is of interest for primary school children who stutter; it might have some relation to social anxiety.

There is an extensive body of research showing that children from different cultures and languages who stutter have higher scores than controls for this test. Children respond true or false to 35 questions that include “people worry about the way I talk,” “my classmates don't think I talk funny,” “my parents like the way I talk,” “I don't mind asking the teacher a question in class,” and “I talk well with most everyone” (p. 73).<sup>203</sup>

The Communication Attitude Test was used to assess 70 stuttering and 271 control 7–14 year old Belgian children.<sup>204</sup> From age 7 years the children who stuttered had more negative attitudes to communication than peers. Additionally, their communication attitude worsened with time but became healthier with the controls. Other reports with 143<sup>205</sup> and 110<sup>203</sup> Belgian school-age children have replicated those findings.

Those Communication Attitude Test differences have been replicated with Italian 7–14 year olds<sup>206</sup> and Croatian 7–13 year olds.<sup>207</sup> Eighty stuttering and 80 control Japanese school-age children, ages 5–12, showed differences,<sup>208</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>209</sup> and Slovenian<sup>210</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>211</sup> The study that reported about communication apprehension<sup>179</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 those significant results<sup>212</sup> with adolescents 13–18 years: a group of 53 receiving stuttering treatment (mean age 15.2 years) and 53 controls (mean age 14.8 years). The study of 36 adolescents mentioned previously<sup>180</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>213</sup>

*Psychological distress*

A birth cohort study<sup>214</sup> identified 217 adolescents who were stuttering at 16 years according to their parents, 137 of whom completed the Rutter Malaise Inventory.<sup>215</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>214</sup> The clinical history of the participants was not reported. Results showed that the stuttering adolescents 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>216</sup> and another<sup>217</sup> has reported the opposite effect for 54 participants and controls.

*Peer awareness*

The earliest report that school-age children are aware of stuttering in peers was in 1958 with 120 children 5–8 years old.<sup>218</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>219</sup> In that 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>220</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>221</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>222</sup> The children rated the videos for a range of attributes dealing with “themes of peer friendship, listener comfort, and allowing a peer who stutter to take a speaking role in a group project” (p. 208). 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>223</sup> However, a report<sup>224</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, bullying during the school years is strongly associated with anxiety later in life,<sup>225,226,227,228,229,230</sup> and one report shows this effect concurrently for a control and a group of adults with stuttering.<sup>231</sup> Hence the association of bullying with stuttering school-age children is of interest in the present context. One report<sup>175</sup> showed stuttering school-age children to have a 63% risk of bullying compared to 22% for controls. Another report<sup>232</sup> of 28 children who stuttered, 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. A report<sup>180</sup> of 36 adolescents indicated that 63% reported bullying 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>212</sup> age 13–18 years, with a mean age of 15.2 years, more adolescents reported bullying: 43% compared to 11% for controls. The study discussed earlier,<sup>190</sup> with 75 stuttering and 150 control children, indicated higher scores for the children who stuttered on the Culture Bullying scale of the Personal Experiences Checklist Child Report.<sup>233</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>217</sup> indicated significantly more bullying for the former group using the The Bully–Victimization Scale.<sup>234</sup>

Another report<sup>235</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 children who stuttered were placed in that category but 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>236</sup> with a mean age of 10 years, used the Peer Attitudes Toward Children Who Stutter scale. It found that children who had contact with a stuttering child had significantly more negative attitudes to children who stuttered. The same research group replicated that finding with 760 children ages 6–12 years.<sup>237</sup>

Retrospective reports of bullying by adults have produced results consistent with the above findings. In one report,<sup>238</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 bullying, and 46% reported long-term effects. These results were replicated with a survey of 324 adult respondents,<sup>239</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>240</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>241</sup>

### *Quality of life*

Those findings about bullying and mental health with stuttering school-age children were consistent with a report using the OASES quality of life measure for 50 stuttering 8–11 year-olds and controls.<sup>242</sup> The children who stuttered had significantly lower quality of life 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.

## **Conclusions**

In contrast to pre-school children who stutter, there is much more direct 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 worsen during this period, with findings of problem anxiety measures more typical of older participants in studies. Two reports<sup>182,190</sup> have found evidence during the primary school years and adolescence of the diagnosable anxiety related mental health disorders that trouble adults. The latter report found evidence that 24% of school-age children presenting at speech clinics for stuttering treatment were diagnosed with social anxiety disorder. The former report contained evidence of worsening anxiety test scores during adolescence. Evidence of bullying during the school years, and negative classroom experiences, are consistent with those findings. The most

prominent anxiety disorder with adults who stutter—social anxiety disorder—is typically diagnosed during early adolescence,<sup>243,244</sup> with median onset at 13 years.<sup>32</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 generally are at risk for developing mental health problems, many of them involving anxiety.<sup>245,246,247</sup> The latter report was of 258 five-year-olds who were diagnosed with a speech or language disorder (only five were diagnosed with stuttering).<sup>248</sup> 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>247</sup> That could certainly be said of stuttering.

### **STUTTERING, MENTAL HEALTH, AND THE TIMING OF EARLY INTERVENTION**

Early intervention is by far the best clinical option for the disorder, as outlined during Lectures Six and Seven. Considering epidemiological data and evidence of the potential quality of life impairment from chronic stuttering, and the mental health evidence presented during this lecture, the following policy statement about the timing of early intervention seems inevitable:<sup>249</sup>

Stuttering typically starts during the pre-school years and is a significant risk factor for 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 recover naturally from stuttering, recovery rate during the first 18 months is estimated to be only 6–8%. 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.

### **SUMMARY**

Adults who stutter presenting at speech clinics may 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. Clinicians need to be mindful of the possibility that techniques for stuttering control may be safety behaviours that sustain speech-related anxiety. Primary school age and adolescent clients seeking stuttering treatment are more likely than younger clients to experience clinically significant anxiety. There is evidence that the psychological problems associated with stuttering begin early during life. Consequently, after diagnosis, early stuttering should be treated with an appropriate evidence-based treatment as soon as possible.

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## **LECTURE ELEVEN**

### **TREATMENT OF SOCIAL ANXIETY<sup>†</sup>**

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#### **SPEECH-LANGUAGE PATHOLOGISTS AND ANXIETY TREATMENT**

As noted during the previous lecture, 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 being said, not all clinicians will have the necessary training or experience to manage social anxiety with stuttering clients. Treatment of anxiety is fundamentally 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 professional body. (p. 195–196)

#### **ANXIETY MEASUREMENT FOR SPEECH-LANGUAGE PATHOLOGISTS**

##### **Background**

It is important to stress that 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. As noted during the previous lecture, adults seeking treatment for stuttering often are affected by social anxiety disorder. An overview is available of clinical measures for that specific disorder that clinical psychologists can use.<sup>3</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.

A specific caveat is needed for speech-language pathologists about measuring the anxiety of children, because it is a lot different to measuring anxiety with adults. One complicating factor is the possible limitations 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. It would be prudent for speech-language pathologists only to screen children for anxiety to determine whether referral to a clinical psychologist is necessary.

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<sup>†</sup> Thanks to Ross Menzies and Lisa Iverach for guidance with this material.

## The Unhelpful Thoughts and Beliefs About Stuttering (UTBAS) scales

### *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,<sup>4</sup> and is presented in Appendix One of this lecture. A Japanese version of the scale is available.<sup>5</sup>

Clinical psychologists and speech-language pathologists developed the scale<sup>6,7</sup> by producing a list of 66 commonly occurring unhelpful thoughts about stuttering expressed by those who stutter. To complete the scale, for each of those thoughts the client indicates how frequently it occurs on a scale of 1 to 5: 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.

### *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" 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 for clients. That may not be so advisable with younger adolescents, and the scale is probably of limited use with school-age children.

### *Interpreting UTBAS scores*

The table shows data for 140 adult stuttering participants:<sup>7</sup> means and standard deviations for UTBAS I, II and III, and UTBAS total score for participants who did and did not have a DSM-IV social anxiety disorder diagnosis. The table has bolded mean UTBAS I scores. The table also shows the results for those who did and did not meet an ICD-10<sup>8</sup> first-stage screening for anxious personality disorder. The mean score differences between participants with and without an anxiety disorder are presented also. All UTBAS scores for those with an anxiety diagnosis are higher than scores for those without an anxiety diagnosis.

	% N	UTBAS I		UTBAS II		UTBAS III		TOTAL	
		MEAN	SD	MEAN	SD	MEAN	SD	MEAN	SD
<i>Social anxiety disorder</i>	24 32	<b>198</b>	59	176	50	201	59	577	153
<i>No social anxiety disorder</i>	77 104	<b>154</b>	49	135	41	146	58	435	150
<i>Mean difference</i>		<b>45</b>		41		54			142
<i>Anxious personality disorder</i>	30 42	<b>208</b>	44	187	51	210	54	605	141
<i>No anxious personality disorder</i>	70 98	<b>146</b>	44	128	43	138	41	413	128
<i>Mean difference</i>		<b>62</b>		59		72			193

### The UTBAS-6

There is a six-item screening version of the UTBAS, known as the UTBAS-6,<sup>9</sup> which is presented in Appendix Two of this lecture. The six items are able to 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. 18).<sup>9</sup>

These are the test items, 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

The diagram shows an example of an UTBAS-6 form completed by a client with clinically significant anxiety.

**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.																
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

**The Fear of Negative Evaluation (FNE) scale***The original 30-item FNE scale*

The FNE scale was published originally in 1969<sup>10</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>7,11,12,13,14,15</sup>

*The Brief FNE scale*

Subsequent to 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>16,17,18,19,20,21</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.

Each of the eight items is scored on a scale of 0–4: 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*. Those numerical scores are assigned to each item and then summed. The range of scores will therefore be 0–32.

These are the test items:

- (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 16 and 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 for adults 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 One). One report<sup>18</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, that table suggests a cut-off score of 25 for potentially clinically significant anxiety. That 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.

That report<sup>18</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) 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)**

Previous measures discussed have dealt with trait anxiety, which is anxiety linked to temperament. However, the SUDS measures state anxiety, which are immediate emotional responses to everyday experiences. The SUDS is usually attributed to the psychologist Wolpe during the 1960s. 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 with 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 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. That 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<sup>23</sup> is presented in the diagram.



### **The Spence Children's Anxiety Scale**

#### *Overview*

The Spence Children's Anxiety Scale has been shown to be reliable and valid, and has comprehensive normative data available. The scale is well established, extensively used, accessible from a website without charge<sup>24</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>25</sup> the former containing 45 items and the latter 38 items. Responses to items are scored on a four point scale: 0 = *never*, 1 = *sometimes*, 2 = *often*, 3 = *always*. 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.” There are subscales 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 less than 10 above the mean is not considered to be concerning. The scale is not intended as a standalone diagnostic instrument:<sup>25</sup>

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.

However, speech-language pathologists could use the parent report scale for screening purposes to determine any need for a clinical psychology referral. It is not advisable for a speech-language pathologist to use scale that requires child responses. Nor is it advisable for a speech-language pathologist to use the subscales, only the total score as a screening measure.

*Pre-school version (3–5 years)*

The scales have a version for 3–5 year-olds<sup>26</sup> comprising 28 parent report items that provide an overall anxiety measure plus subscales that deal with specific aspects of child anxiety: “generalized anxiety, social anxiety, obsessive compulsive disorder, physical injury fears and separation anxiety.” 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 follow instructions, which can be downloaded from the website.<sup>24</sup> Parents respond with a 5-point scale, indicating the extent to which each of the 28 statements pertain to their children: 0 = *not true at all*, 1 = *seldom true*, 2 = *sometimes true*, 3 = *quite often true*, and 4 = *very often true*.

A numerical score is obtained from these responses, and T-scores are available for the subscales and the total score. As was 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**

The Preschool Anxiety Scale Revised<sup>27,28</sup> is a parent-report anxiety measure of anxiety for children younger than 6 years. It consists of 28 items that load onto four subscales: 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”). For each item, parents select the response that best describes the child on a 5-point scale: 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.

The Preschool Anxiety Scale Revised is publicly available in English and seven other languages. Normative data are provided<sup>27</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>27</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>29</sup> shows thousands of publications dealing with the method. A tutorial<sup>1</sup> about CBT for anxiety with stuttering overviews the four standard components of the treatment with specific reference to stuttering: exposure, behavioural experiments, cognitive restructuring, and attentional training. Cognitive Behaviour Therapy is overviewed at this website.<sup>30</sup>

There has been evidence from the 1970s that anxiety treatments such as desensitisation and meditation may benefit those who stutter,<sup>31,32</sup> and it is common to incorporate CBT, or components of it, within speech restructuring treatments.<sup>33,34,35,36,37,38</sup> However, there have been only two conference reports of the value of the CBT component itself,<sup>39,40</sup> and only three published clinical trials that conform to the definition of a clinical trial outlined during Lecture Five.

### CBT for stuttering: A clinical randomised controlled trial

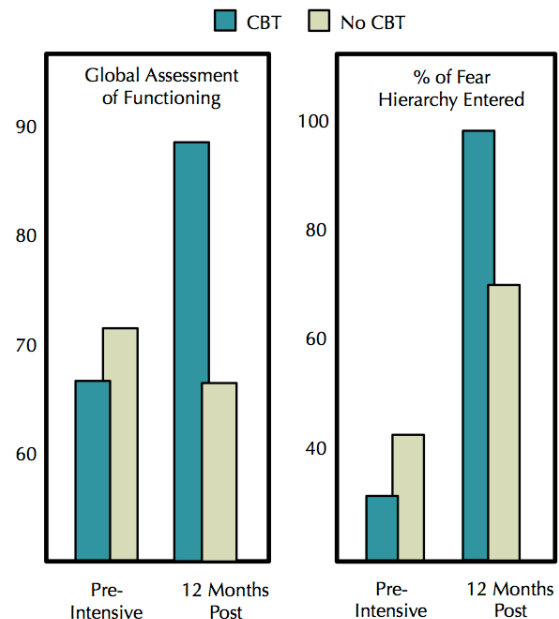
#### Design

The first of those trials<sup>41</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, presented with 10 weekly sessions. Eight sessions were 1 hour, one was 2 hours, and two were 3 hours. There were seven participant drop-outs (22%).

#### Results

The trial showed clearly 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. This DSM-IV index is a score out of 100 indicating general mental health and wellbeing and the 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>42</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 normal 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. The difference between the groups was significant at 12 months post-treatment. These results are presented in the figure.

## Development of standalone Internet CBT for stuttering

### *Clinical issues driving the development*

Despite its promise, the clinical trial just described raises various clinical issues.<sup>43</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, that raises the matter of whether, on the whole, such speech-language pathology interventions will be as effective as those provided by clinical psychologists. It would be difficult to argue that 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 those 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>43</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>44</sup>

There are good reasons to foreshadow that a solution to this problem can be an interactive, Internet-driven CBT treatment that requires no clinician. Speech-language pathologists would be able to integrate CBT treatment with speech restructuring treatment without needing psychological training or access to a clinical psychologist, and be able to do so in a cost neutral manner.

A review<sup>45</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 argued also that human therapist simulation would require corrective feedback for incorrect client responses during the learning process of CBT, and 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, so that users receive “a large number of opportunities to engage in cognitive and behavioural tasks relevant to their problems” (p. 251).<sup>45</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 those guidelines, the authors designed a standalone Internet CBT treatment: CBTpsych.<sup>46</sup> The program design was based on the original clinic treatment<sup>41</sup> and incorporated components of the Clark and Wells social anxiety model described during the previous lecture.<sup>47,48</sup> The program incorporates the faces and voices of a man and woman clinical psychologist who communicate to the user throughout the treatment. The treatment is designed around an on-line pre-treatment assessment given by CBTpsych: the 30-item FNE, UTBAS and the Depression Anxiety Stress Scales.<sup>49</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 CBTpsych. 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 are able to control their thoughts, and hence control emotions. Users are shown how both negative and positive thoughts and emotions may emerge from the same event. The example provided is of missing a bus because it departs ahead of schedule. The figure shows how that event may promote thoughts that lead either to positive or negative emotions.

The CBTpsych clinical psychologist then presents a “thinking exercise” where the user is presented with a life situation that does not involve speech and is required to write different thoughts about the situation that could lead to the different emotions of anxiety, anger, sadness and happiness. Then the program compares the user responses with the responses prepared by the clinical psychologist.



**I'm sick of this bus company  
The driver must be incompetent  
I don't deserve this**

Then 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 CBTpsych responses. One response given 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 the response given that would lead to happiness is “I only had to repeat once what I was asking for.” Similar thinking exercises recur throughout Section One.

**I have an excuse for being late  
I have an hour for coffee  
I can read my book**



CBTpsych 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 in to 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. CBTpsych presents examples of such causal thoughts.

The program then explains common cognitive errors, otherwise known as cognitive distortions, such as those outlined in the following table. These are typically incorporated within CBT.<sup>50</sup> CBTpsych then presents numerous examples of such 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 requires the user to identify which of the cognitive errors they are.

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<i>MIND READING</i>	<i>Assuming people are thinking negative thoughts about you when there is no real evidence that they are.</i>
<i>FORTUNE TELLING</i>	<i>Arbitrarily predicting that things will turn out badly.</i>
<i>EMOTIONAL REASONING</i>	<i>The way you feel about yourself is reality: "I feel stupid so I must be stupid."</i>
<i>MENTAL FILTERING</i>	<i>Dwelling on the negatives and discounting the positives.</i>
<i>"SHOULD" THINKING</i>	<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>
<i>OVERGENERALISATION</i>	<i>Interpreting negative events as part of a never ending pattern of defeat.</i>
<i>ALL OR NOTHING THINKING</i>	<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>
<i>DISCOUNTING THE POSITIVES</i>	<i>Belief that positive achievements don't count in evaluating yourself.</i>
<i>LABELLING</i>	<i>Thinking "I am a loser" Instead of "I made a mistake."</i>
<i>PERSONALISATION</i>	<i>Blaming yourself for something that was not entirely your fault.</i>
<i>BLAME</i>	<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. Those standard cognitive challenges are: (p. 264–265)<sup>46</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 each of them, CBTpsych has cognitive restructuring sample answers for each of the eight probe questions in the table, totalling 528 sample answers. In order to ensure an adequate dose of cognitive restructuring, CBTpsych requires users to write at least 40 different

restructurings of their unhelpful thoughts and beliefs. For each cognitive challenge that the user writes, CBTpsych provides from its database a predetermined challenge for comparison. For example, in response to the unhelpful thought "It's impossible to be successful if you stutter," this was a user response to the probe question "what would you tell a friend, to help, if he/she had the thought?" (p. 264):<sup>46</sup>

It is absolutely wrong. You can be successful in many things despite you [sic] stutter, look at your past performance, you succeed sometime.

The CBTpsych sample answer was:

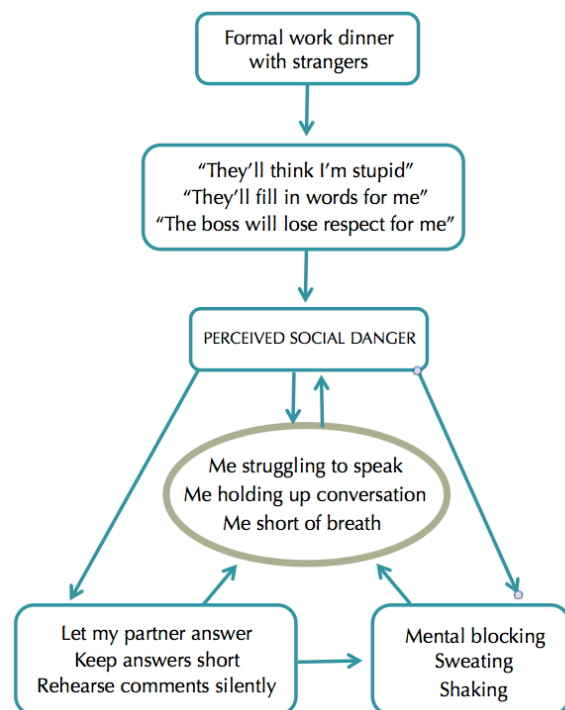
Don't be silly! Lots of people who stutter are successful. This thought is so self-defeating. You need to beat it!

### 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. Then, users are guided in building 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 during the program to establish behavioural experiments and to target unhelpful imagery for correction. Here is an example of an individual formulation that might occur for a user.<sup>‡</sup> It concerns the social event of a formal work dinner with strangers.

That 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." Those 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 feeling of self consciousness, which feeds into a cycle



<sup>‡</sup> 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.

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 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 CBTpsych clinical psychologists says this to 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 designed to test out your negative thoughts in situations where we will ask you to drop your avoidance and your safety behaviours. 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, CBTpsych 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 users select 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. CBTpsych 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 situations.

CBTpsych 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 during Section Three, during behavioural experiments. The following is an example of a behavioural experiment provided by CBTpsych 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, in writing about "why it doesn't matter what other people think of me." One of the CBTpsych 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 CBTpsych 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.

CBTpsych 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 so distasteful. The CBTpsych 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."



CBTpsych 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 in exploring 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, CBTpsych 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>51</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 CBTpsych, on which the psychologist's voice provides training in shifting attention rapidly from one focus to another. When the user has practiced the attentional training technique daily for some weeks and mastered it, the CBTpsych 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.

CBTpsych then presents users with a list of their commonly avoided everyday situations scored at the online pre-treatment assessment with the Stuttering Specific Avoidance Scale. The user is then required to choose three of them and to practice the situational attentional refocusing technique in each of them and to record what occurred during each practice. The user is urged to continually practice the technique.

The final part of Section Six deals with problems of mental imagery that involve the biased observer perspective that often affects those with social anxiety, as discussed during Lecture Ten:

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.

CBTpsych presents an example of a social encounter during which stuttering occurs. The images of that encounter recur for the person two years later. The woman recalls seeing herself during the event looking anxious and tense and those present evaluating her quizzically and apprehensively, and generally having a negative view of her, as shown in the image on the left.



However, that recall of the situation cannot be correct, and what she actually saw—the field perspective—was more like the image on the right. Yet, years later, she still ruminates about how badly she thought the social encounter went and the thought of it all makes her ruminate about how badly future social encounters will turn out and makes her anxious about them before they even occur. The CBTpsych program invites users to test whether this is an issue for them:

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>52</sup> for faulty images of past events such as in the previous example. Users download an audio file from CBTpsych, on which a

psychologist's voice provides guidance in mentally going over a past event and re-scripting it so that it is different from the troublesome version; 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, and emphasises that minor setbacks are inevitable, and should not be interpreted as relapse. The clinical psychologist guides users in recognising 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.

### **Phase I clinical trials of CBTpsych**

#### *Background*

The developers reported preliminary data for two adult participants.<sup>46</sup> Participant 1 completed the treatment in 2 months with 11 log-ins and Participant 2 completed the treatment in 3.5 months with 34 log-ins. Results suggested that treatment effects with this intervention may be similar to those attained with the clinic version that was presented by a clinical psychologist.<sup>41</sup> At post-treatment, both participants no longer had social anxiety disorder diagnoses, and various anxiety measures showed improvements.

#### *Method*

Subsequently, the prototype of the website was refined and a Phase I clinical trial was reported<sup>43</sup> with 19 adult participants recruited from a speech-language pathology clinic waiting list. Five of those (26%) did not begin the treatment, leaving 14 participants, who were permitted 5 months to complete the treatment. Their average age was 42 years (range 33–77 years). Several psychometric assessments were collected pre-treatment and at 5 months after the start of the treatment, independently of the measures collected by CBTpsych.

The presence or absence of social anxiety disorder was assessed with a standard, self-administered computer assessment.<sup>53</sup> Additionally, participants completed the 30-item FNE scale and the UTBAS scale, the Beck Depression Inventory,<sup>54</sup> the State-Trait Anxiety Inventory,<sup>55</sup> the Social Phobia Anxiety Inventory,<sup>42</sup> and the Endler Multidimensional Anxiety Scales-Trait.<sup>56</sup> Quality of life was measured with the OASES and stuttering severity was measured with %SS from two unscheduled 10-minute telephone calls from strangers.

#### *Results*

Users had a mean of 15 log-ins with the mean log-in time of 7 hours and mean period between log-ins of 7 days. However, as outlined earlier, much of the CBTpsych clinical procedures occur during everyday situations when users are not logged in. Eight of the 14 participants completed all seven CBTpsych sections during the 5-month access period. Users did not have any contact with a clinical psychologist or a speech-language pathologist during the trial.

At pre-treatment, seven participants met diagnostic criteria for social anxiety disorder, and at post-treatment only two retained that diagnosis. However, those two participants did not complete the entire CBTpsych treatment, one progressing only to Section Two and the other to Section Four. Two of the four psychometric measures showed statistically significant improvement at post-treatment: the FNE scale and the Social Phobia Anxiety Inventory. UTBAS scores showed significant improvement, and OASES total scores and quality of life scores showed significant improvement. Percentage syllables stuttered scores showed no change from pre-treatment to post-treatment.



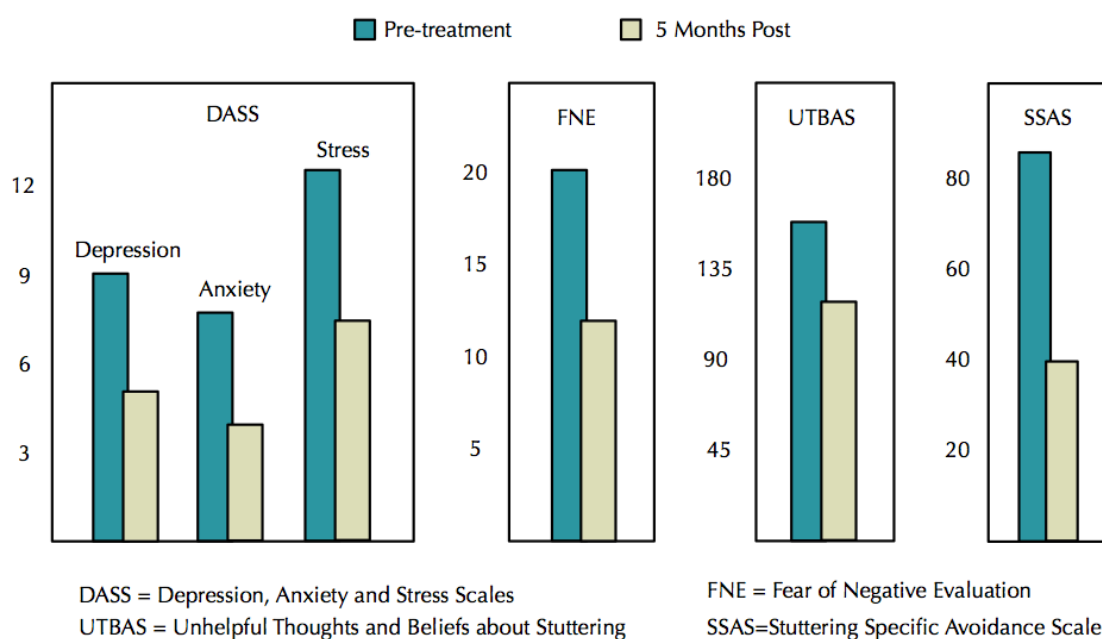
## A Phase II clinical trial of CBTPsych

### Background

In formulating a Phase II trial of this standalone Internet treatment,<sup>57</sup> the developers noted that 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 CBTPsych. 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 49 of 267 participants (18.4%) and the completion rate for CBTPsych was 52 of 267 participants (19.5%). This completion rate was far superior to existing standalone Internet treatments for depression and anxiety, which attain below 7%<sup>58</sup> and around 1%.<sup>59</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 CBTPsych and trials of in-clinic CBT for stuttering with a clinical psychologist. Post-treatment scores for the Depression, Anxiety and Stress Scale were within normal community values. Results are shown in the figure.<sup>‡</sup>

<sup>‡</sup> 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.

### Acceptance and commitment therapy

Subsequent to 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>60</sup> One of those is Acceptance and Commitment Therapy, commonly known as ACT. A user guide to ACT is available.<sup>61</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>62</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 “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>63</sup> The overall purpose of ACT is<sup>60</sup>

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)

A recent review of the efficacy of ACT was<sup>64</sup> a meta-analysis of 18 randomised controlled trials (N=917). The authors concluded that the treatment was promising, being superior to control conditions. However, there was no evidence of it being superior to established treatments. Another review around the same time,<sup>65</sup> in the context of a general review of third wave therapies, was more guarded, reviewing 13 randomised controlled trials and drawing attention to methodological problems with them, and noting a moderate effect size for ACT. A more recent review by the same author<sup>66</sup> was a meta-analysis of 60 randomised controlled trials, and reported no methodological improvements in trial quality and a reduction to a small effect size.

There has been a description of how ACT may pertain to those who stutter,<sup>67</sup> and a subsequent report of an ACT package tailored specifically for stuttering.<sup>68</sup> Twenty 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 treatment rather than the ACT treatment. The speech treatment was described as “fluency shaping activities, speech rate control, speech naturalness and self-administered timeout for stuttered moments” (p. 291).<sup>68</sup> Results at 3 months follow-up showed statistically significant improvements for stuttering severity during speech in 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.

Another pertinent report<sup>69</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 improvement across a range of psychological measures. However, no data were presented to suggest that the addition of mindfulness training improved the effects of CBT.

### 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 typically is not a primary professional domain. However, there are anxiety measurement procedures suitable for speech-language pathologists, who may wish to provide anxiety treatment with 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

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<sup>†</sup> The attentional training in Section Six of CBTpsych described earlier is a mindfulness procedure.

that a standalone Internet social anxiety treatment is efficacious. Hence, speech-language pathologists might recommend it for their clients without cost or professional training. This could prove to be a significant advance for speech-language pathologists who do not have professional qualifications for anxiety management.

## APPENDIX ONE

### THE UNHELPFUL THOUGHTS AND BELIEFS ABOUT STUTTERING (UTBAS) SCALE

#### Unhelpful Thoughts and Beliefs About Stuttering Scales (UTBAS I, II, III) For Adults

		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	People will doubt my ability because I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
2	It's impossible to be really successful in life if you stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
3	I won't be able to keep a job if I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
4	It's all my fault – I should be able to control my stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
5	I'm a weak person because I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
6	No one will like me if I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
7	I might stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
8	People focus on every word I say	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
9	I am incompetent	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
10	No one could love a stutterer	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
11	I will stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
12	Everyone in the room will hear me stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
13	I'm stupid	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
14	People will think I'm stupid if I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
15	I'll never be successful because of my stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
16	I won't be able to answer their questions	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5

Using the following scale, please read each item below and circle the number which most accurately describes you in terms of:  
 (I) how frequently you have these thoughts, (2) how much you believe these thoughts, (3) how anxious these thoughts make 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

	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				
17	I'm hopeless	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
18	I'm of no use in the workplace	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
19	People will think I'm incompetent because I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
20	I'll block completely and won't be able to talk	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
21	Everyone will think I'm an idiot	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
22	I can't speak to people in positions of authority	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
23	People will think I'm strange	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
24	People will think I can't speak English	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
25	No one would want to have a relationship with a stuturer	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
26	I can't think clearly because I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
27	I can't speak to aggressive people	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
28	People will think that I have no opinions	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
29	People will think I'm boring because I have nothing to say	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
30	If I block, people will think I'm retarded	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
31	I can't face these people	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
32	People will wonder what's wrong with me if I stutter	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
33	What will people think of me if they disagree with what I say?	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
34	Most people view stutters as less capable	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
35	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

	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	2	3	4	5	1	2	3	4	5	1	2	3	4	5
36	My pauses are too long – people will think I'm weird															
37	People won't like me because I won't be able to talk															
38	I can't convince people of anything I say because I stutter															
39	People will think I'm retarded if I stutter															
40	I'll block – I know I will															
41	I'll make a fool of myself															
42	People get tired of waiting for me to get my words out															
43	People shouldn't have to wait so long for me to speak															
44	I always embarrass the people I'm speaking to															
45	People think I have something to hide because my stutter sounds suspicious															
46	People will think that I'm worthless															
47	I'll embarrass myself															
48	I can't speak to people I find sexually attractive															
49	No one will understand what I'm trying to say															
50	What's the point of even trying to speak – it never comes out right															
51	I won't be able to say exactly what I want to say															
52	Everyone will think I'm simple or dumb because I avoid using difficult words															
53	I slow up everyone's conversation															
54	Everyone hates it when I start to speak															

	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	2	3	4	5	1	2	3	4	5	1	2	3	4	5
55	I can never speak on the phone	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
56	I won't be able to ask for what I want	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
57	The person on the other end of the phone will hang up on me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
58	People will laugh at me	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
59	People will think I'm mute	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
60	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
61	The answering machine will turn off if I block – I won't be able to leave any message	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
62	They'll think I'm a prank caller if I block	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
63	I won't be able to say 'hello' when I pick up the phone	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
64	People who stutter are stupid	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
65	People who stutter are incompetent	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
66	People who stutter are boring	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5

**APPENDIX TWO  
THE UTBAS-6 SCALE**

**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.

	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	2	3	4	5	1	2	3	4	5	1	2	3	4	5
1	I'll never be successful because of my stutter															
2	People will think I'm incompetent because I stutter															
3	People will think I'm strange															
4	I don't want to go – people won't like me															
5	What's the point of even trying to speak – it never comes out right															
6	I'll never finish explaining my point – they'll misunderstand me															

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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