

The University of Newcastle

Treatment Schedules in the Delivery of the Lidcombe Program of Early Stuttering Intervention

A thesis submitted in fulfilment of the requirements for
the degree of Doctor of Philosophy in Speech Language
Pathology

Sarita Koushik, MSLP (C), BSc.

School of Humanities and Social Sciences
Newcastle, NSW

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STATEMENT OF ORIGINALITY

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Endorsement by Supervisor:

Sally Hewat, PhD

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DEDICATION

This thesis is dedicated to my father Raj Koushik (1938-2000). He would be proud to know that his two daughters achieved their goals by following his example of hard work, commitment and perseverance.

PUBLICATIONS / CONFERENCE PRESENTATIONS / POSTERS RESULTING FROM THIS THESIS

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LIST OF ACRONYMS USED IN THESIS

ADHD	Attention deficit hyperactivity disorder
AQS	Attachment Q-Set
ASRC	Australian Stuttering Research Centre
BC	Beyond-clinic
CBCL	Child Behaviour Checklist
CEBM	Oxford Centre for Evidence-Based Medicine
CSP	Comprehensive Stuttering Program
DCM	Demands Capacities Model
EMG	Electromyography
EBP	Evidence-based practice
FRP	Fluency Rules Program
HREC	Human Research Ethics Committee
ICC	Intra-class correlation
ISTAR	Institute for Stuttering Treatment and Research
LPTC	Lidcombe Program Trainer's Consortium
MLU	Mean length of utterance
NHMRC	National Health and Medical Research Council
PCI	Parent-child interaction
RCT	Randomised controlled trial
SI	Syllable initiation
SR	Severity rating
%SS	Percent syllables stuttered
STS	Syllable timed speech
UofN	University of Newcastle
VRCS	Verbal response contingent stimulation
WC	Within-clinic

PRÉCIS

Chapter 1 presents an overview of stuttering including, the 1) characteristics, 2) cause, 3) development, 4) effects of natural recovery, 5) treatment, and 6) impact of the disorder. The information presented in this chapter gives sound rationale for the questions asked in the empirical studies in the thesis. The chapter concludes that early intervention is essential. With regard to evidence-based practice (EBP), Dollaghan (2007) suggested that EBP is the “the conscientious, explicit and judicious integration of 1) the best available external evidence from systematic research, 2) best available evidence internal to clinical practice, 3) best available evidence concerning the preferences of a fully informed patient” (p. 2). Dollaghan considers all 3 components of this definition as equally important. In this thesis, external evidence is explored to determine the best available evidence from systematic research. The methods of Onslow and colleagues’ (2008) and NHMRC’s (2009) guidelines for evaluating the current level of evidence will be used in the review of early stuttering treatment outcome reports.

In Chapter 2, the research evidence from early stuttering reports is categorised according to the theoretical framework, including multifactorial models, speech restructuring models and verbal response contingent stimulation. The conclusion formed in this chapter is that the Lidcombe Program has the largest available clinical trials evidence and highest level of evidence according to NHMRC (2009) guidelines. The Lidcombe Program is the treatment option chosen for the two empirical studies presented in this thesis.

Chapter 3 presents all other Lidcombe Program evidence as clinicians draw on related research for best clinical practice. In the presentation of this evidence, a gap in the literature was found in relation to two file audit studies performed in Australia and the United Kingdom (Jones et al., 2000; Kingston et al., 2003). These studies examined

the relationship between predictor variables and the number of clinic visits to complete Stage 1. However, the variable, average time between clinic visits, was not explored as a predictor for treatment outcomes.

Chapter 4 presents the first empirical study of this thesis on a North-American file audit of the Lidcombe Program. This study replicates and extends the methodology of Jones et al. (2000). The predictor variable, average time between clinic visits, was included in the methodology. Evidence was found in this study that the Lidcombe Program is not delivered on a weekly basis in clinical communities. However, there is no available evidence as to how altering weekly clinic visits might affect treatment efficacy and efficiency.

Chapter 5 presents the second empirical study, evaluating the effects of varying Stage 1 Lidcombe Program treatment schedules on treatment efficacy and efficiency. This study is a prospective Phase II clinical trial of 3 different service delivery models including weekly, twice weekly and fortnightly treatment. Findings suggest that fortnightly clinic visits might be an efficient and efficacious alternative to weekly Lidcombe Program clinic visits. Chapter 6 presents a discussion of the results of both empirical studies and suggests further areas of research.

ABSTRACT

This thesis presents two empirical studies of the Lidcombe Program of early stuttering intervention.

A retrospective file audit of the Lidcombe Program was performed in North America in order to evaluate the relationship between specific case variables and treatment time during Stage 1 (instatement of fluency). The study was a replication and extension of the file audit study by Jones et al. (2000), with the time between clinic visits being considered as an additional variable. The variables were extracted from files of 138 children younger than 6 years who had completed Stage 1 of the treatment. The results showed that the median number of clinic visits to complete Stage 1 was 11. High pre-treatment stuttering severity predicted more clinic visits. Mean interval between clinic visits of fewer than 11 days was associated with longer treatment times than mean interval of 11 days or more. The results for North America were generally consistent with benchmark data from the United Kingdom and Australia. The data from previous Australian and British studies were combined with the North American data and a meta-analysis was performed to establish worldwide clinical benchmarks. This study indicated the potential clinical significance of attendance schedule and prompted further investigation.

A Phase II prospective clinical trial of different treatment schedules for the Lidcombe Program was conducted to evaluate the effects of treatment schedules during Stage 1 clinic visits, with reference to treatment efficiency and efficacy. Twenty-one children were randomly allocated into one of three Lidcombe Program treatment schedules: attendance weekly, twice weekly, or fortnightly. It was found that the median number of clinic visits to complete Stage 1 by treatment schedule was 23 for weekly attendance, 27 for twice weekly attendance and 10 for fortnightly attendance. The

findings suggest that fortnightly attendance was efficient and efficacious for the children in this study. The implications for an alternative service delivery model with the Lidcombe Program are discussed.

CHAPTER 1

Overview of Stuttering and Evidence-Based Practice

Description of Stuttering

Stuttering is a speech disturbance that involves difficulty producing utterances with a natural flow. The stuttering disturbance can be described as repeated movements, fixed postures and superfluous behaviours (Packman & Onslow, 1998; Teesson, Packman, & Onslow, 2003). Repeated movements may occur on the entire syllable, such as “where, where, where”. Repeated movements may also occur on an incomplete syllable, such as “sa, sa, santa”. The third category of repeated movements may occur on multisyllable units, such as “instru, instru, instrument.”

The second stuttering disturbance is fixed postures. The articulators move continuously during normal speech movements. Fixed postures are a static position of articulators. Fixed postures may occur with or without audible airflow. Fixed postures with audible airflow may sound prolonged, such as “I caaaaaaaan run.” If no audible airflow is present, the speech production may appear “stuck” or “blocked”. The duration of blockage may vary in length.

The third stuttering disturbance is superfluous behaviours. These disturbances can be nonverbal or verbal. Superfluous behaviours are actions that are not intended as part of the original communication. Verbal superfluous behaviours may involve redundant words or phrases, such as “I - well ah uh ah - went to the store.” Nonverbal superfluous behaviours may involve facial contortions or other movements such as raised eyebrows, blinking or stamping feet.

Cause of Stuttering

The cause of stuttering is unknown (Guitar, 2006). However, researchers have developed theories in an attempt to explain the cause. Theories of stuttering have been important in the development of treatment approaches. The sensory-motor modelling theory proposes that people who stutter are deficient in neuronal processing resources (Packman & Attanasio, 2004). These resources are responsible for maintaining the relationship between the auditory and motor systems that underlie speech production. The deficiency may be identified by delayed speech, articulatory errors or deficits in central processing. The neuroscience model proposes that stuttering is caused by instability in the speech motor control system (Packman & Attanasio, 2004). The theory proposes that there are two functional loops, the first controlling cognition and second the programming of sounds. The instability between the functional loops results in stuttering. A more recent report theorises that stuttering is a problem of neural function in the supplementary motor area of the brain (Packman, Code, & Onslow, 2007). The syllable initiation theory proposes that stuttering is a disorder of the initiation of speech motor plans at the level of the syllable (Packman, Code, & Onslow, 2007).

Development of Stuttering

Stuttering generally begins between 2 and 5 years of age, during the preschool years (Yairi & Ambrose, 1999). The onset may be sudden or gradual (Yairi & Ambrose, 1999). A recent large-scale prospective study of 1619 children reported stuttering onset to be rapid and episodic (Reilly, Onslow, Packman, Wake, Bavin, et al., 2009). In that study it was found that factors associated with the onset of stuttering include male gender, twin birth status, high maternal education and a high vocabulary score at 2 years of age.

Estimating the incidence of stuttering during the preschool years is currently controversial. Previous reporting gave the incidence during these years as 5% (Andrews & Harris, 1964; Mansson, 2000). However, those estimates were challenged in a methodologically advanced study (Reilly et al., 2009) of 1619 children under the age of three. That study was the first to employ a community cohort, ascertained prior to the onset of stuttering, with expert diagnosis a short period after onset. One aim of the study was to measure the incidence of stuttering. The three-year cumulative incidence of stuttering was 8.5% (Reilly et al., 2009). The cumulative incidence graph of the data suggested that more cases would occur after 3 years of age. As the cohort continues to be followed, the estimate of preschool incidence is likely to be 2-3 times higher than existing estimates of 5%.

Estimates of prevalence rates vary across countries. Bloodstein and Bernstein Ratner (2008) presented two tables of prevalence rates among schoolchildren in the United States and in other countries. The following summary statistics were prepared by the author of this thesis. The distribution of the overall scores was not normal looking, with two outlying data points. The median values were used because the distribution of scores appeared skewed. The median prevalence rate for school-age children in the United States was 0.82% ($R = 0.30 - 2.00$, $SD = 0.55$). In other countries it was 1.11% ($R = 0.58 - 2.2$, $SD = 1.08$) and combined it was 0.97% ($SD = 0.93$). The median prevalence in other countries was slightly higher than that reported from the United States. The data from these reports are cross-sectional and without expert diagnosis. To date, no prospective cohort estimate of prevalence exists. The absence of prospective studies is a caveat to the presented findings.

Natural Recovery

Many children who begin to stutter recover naturally without receiving treatment (Bloodstein & Bernstein Ratner, 2008). Controversy exists as to the percentage of natural recovery during the preschool years. Mansson (2000) followed over 1000 children from birth to age five and found that 71.6% recovered naturally within 2 years of the study commencing. Other studies indicate a recovery rate for early stuttering of 74% within the first 4 years (Bloodstein & Bernstein Ratner, 2008; Yairi & Ambrose, 1999). Yairi and Ambrose (2005) found that natural recovery was more likely for girls than boys, for children with a shorter time since onset, and for those with a family history of natural recovery. Although the recovery rate appears high, evidence for predictors of natural recovery are not conclusive (Yairi, Ambrose, Paden, & Throneburg, 1996).

Treatment for Stuttering

Onslow and Packman (1999) discussed issues related to when to commence treatment, and which treatment program to employ. In regards to when to commence treatment, clinicians are uncertain if early stuttering should be treated shortly after onset or delayed due to the potential for natural recovery (Yairi & Ambrose, 1999). However, if treatment is delayed due to waiting for natural recovery, the child may be denied an effective treatment. If the decision is made to treat, then the timing of intervention becomes an issue. Further, evidence shows that stuttering becomes less tractable with advanced age (Bloodstein & Bernstein Ratner, 2008). Therefore, clinicians need guidance with the decision of when to treat early stuttering. Treatment choice is another important issue for clinicians to consider. Previously, choice of treatment has been based on expert opinion rather than research evidence (Liamputtong, 2010). However, a

shift from expert opinion to evidence-based practice (EBP) has occurred, changing the way clinicians view treatment options and make clinical decisions.

Impact of Stuttering

Researchers agree that stuttering should be treated early, as it is more tractable in children than in adults (Bloodstein & Bernstein Ratner, 2008; Reilly et al., 2009).

Negative peer responses can occur in preschoolers under 6 years of age. Langevin, Packman and Onslow (2009) followed four 3-4 year old children who stuttered in the school yard and measured peer responses to the stuttering child. Some peers reacted negatively by interrupting, showing confusion, mocking, walking away from or ignoring the child. Persistent stuttering can lead to mental health disorders including social phobia, maladjustment and anxiety disorders (Craig & Calver, 1991; Hayhow, Cray, & Enderby, 2002). Further, stuttering adults are at extreme risk of social phobia (Iverach, Jones, O'Brian, Block, Lincoln, et al., 2009c) and at heightened risk of mood and personality disorders (Iverach, Jones, O'Brian, Block, Lincoln, et al., 2010). An efficacious stuttering treatment in adulthood is impeded by the development of anxiety related disorders (Iverach, Jones, O'Brian, Block, Lincoln, et al., 2009a). To avoid these potential negative consequences, early intervention is essential.

Evaluating Early Stuttering Treatments

Early intervention for stuttering is essential to prevent the negative consequences of advanced stuttering for a person's social, emotional and mental health. Many treatment programs for early stuttering have been established (Onslow & Packman, 1999); but few have scientific evidence as a foundation for clinical practice (Mullen, 2007). In the establishment of scientific evidence, treatments generally undergo different stages of development, beginning in controlled research conditions before reaching general clinical communities. The value of evaluating a new treatment under

controlled conditions is to determine whether it is successful before employing it as best clinical practice.

Robey (2004) developed a five-phase model to classify the developmental stages of a treatment in the field of speech pathology and audiology. The purpose was to develop a structure for organising clinical research with the intention of classifying the strength of scientific evidence. A full description of this model is explained later on in this chapter. This model served as a foundation for other classification systems. Onslow et al. (2008) argued that a clinical trial is the fundamental unit in the evaluation of scientific rigor for stuttering treatment reports. They developed a method for evaluating research in stuttering with the intention of removing the burden of work required for clinicians to engage in evidence-based practice. This will be explained later on in this chapter.

In the next section, evidence-based practice is described as the foundation for this method of evaluating research evidence in three areas, namely external systematic evidence, individual clinical expertise and client perspective. A method for evaluating systematic evidence for stuttering research is derived from Onslow and colleagues' (2008) definition of a clinical trial and described in this chapter. Application of this method is seen in Chapter 2 of this thesis.

Evidence-Based Practice

Evidence-based practice was first described by Sackett, Rosenberg, Gray, Haynes, and Richardson (1996) in the field of medicine to classify the quality of research reports. Their definition of EBP highlights the importance of evidence obtained from research, clinical practice and patient perspective. However, the original emphasis was placed on scientific evidence rather than the other two areas (Dollaghan, 2007). Since then, the definition has been applied to other disciplines, including speech

pathology. Dollaghan (2007) expanded the definition to highlight all three aspects of the definition equally. Dollaghan suggested that EBP is the “the conscientious, explicit and judicious integration of 1) the best available “external” evidence from systematic research, 2) best available evidence “internal” to clinical practice, 3) best available evidence concerning the preferences of a fully informed patient” (p. 2). Generally, clinicians who engage in good quality EBP integrate all three levels equally in their clinical decisions.

External Evidence

A comprehensive system for evaluating systematic research is important in the assessment of external evidence. The Oxford Centre for Evidence-Based Medicine (CEBM, 2010) developed a comprehensive system for rating external research evidence. Another hierarchical system developed by the National Health and Medical Research Council (NHMRC) in Australia evaluates levels of evidence, with each level related to research design features, as follows (NHMRC, 2009): Systematic review of RCTs (Level 1), RCTs (Level II), pseudo-randomised controlled trials (Level III-1), non-randomised controlled trials (Level III-2), uncontrolled trials (Level III-3) and case studies (Level IV). The levels of evidence hierarchy is available in Table 1.1.

Table 1.1: Levels of evidence hierarchy by NHMRC (2009) guidelines

Level	Research Design	Ranking
I	Systematic Review of RCTs	Highest Level
II	RCTs	
III-1	Pseudo-Randomised Controlled Trials	
III-2	Non-Randomised Controlled Trials	
III-3	Uncontrolled Trials	Lowest Level
IV	Case Study	

In RCTs (Level II), participants are randomly allocated to experimental groups receiving different interventions. Randomisation reduces bias in a research design and is therefore highly ranked. According to the NHMRC (2009) guidelines, the highest level

of evidence is a systematic review of all randomised controlled studies (Level 1), and the lowest level is a single subject case study (Level IV). Systematic reviews are ranked highest as this research methodology evaluates several different randomised controlled studies. On the other hand, case studies are ranked lowest as only a few participants are involved and the methodology usually trials a new treatment. In pseudo-randomised controlled trials (Level III-1), assignment to an experimental or control group is not fully random. For example, assignment may occur by alternately allocating subjects to a group. This research methodology is therefore less rigorous than randomisation, due to the systematic method for assigning subjects to a group. In non-randomised trials (Level III-2), assignment to a group is determined in a non-random way, for example by clinic site location, whereby a subject is placed in a group because of proximity to a clinic rather than random assignment. Finally, in uncontrolled trials (Level III-3) subjects are assigned to a treatment group and followed, with no comparison to a control group. Results of such a study neither constitute evidence that a treatment works nor indicate how much improvement was made, since no control group was present to allow comparison of research findings.

Research design is important in the evaluation of treatment reports. However, it does not guarantee the quality of the study, so it is important for readers to critically appraise the validity of the report (Liamputtong, 2010). Quality of the study refers to the methodological soundness of the study design. In stuttering research, many researchers have provided recommendations for the quality and relevance of research evidence (Bloodstein & Bernstein Ratner, 2008; Ingham & Riley, 1998). Bloodstein and Bernstein Ratner (2008) outlined a useful set of criteria for evaluating the quality of treatment reports based on empirical evidence. Some of those criteria are: attention to effective methodology with representative participants rather than only to research

design, use of objective measurements such as frequency of stuttering, rate of speech or severity ratings judged by blinded observers rather than experimenters. Long-term follow up investigations are considered important to measure the stability of the treatment and thus the treatment quality. Further, the method must be effective for any qualified clinician, not only experts in the field. In the evaluation of stuttering treatment reports it is important to consider the methodological quality along with systematic evidence from the research design.

Internal Evidence

The second component of EBP refers to the internal evidence obtained from clinical practice (Dollaghan, 2007). This can be explored by well-developed single-subject methodologies or case studies. In this study design, a treatment approach is delivered to a few individuals and their responses to treatment are recorded over time (Liamputtong, 2010). The benefit of recording treatment responses is that clinicians can document outcomes and compare them to pre-treatment measures, thus providing internal evidence for clinical practice. However, the measures can be applied to that individual only and cannot be generalised to the population. Strong evidence internal to the practice must be documented to obtain the best measures of clinical performance (Dollaghan, 2007). Case studies showing clinical promise are highly important for the next stage of development of the treatment. If a treatment proves effective with a small number of subjects it warrants replication with more subjects, providing outcome measures for larger numbers, allowing the efficacy of the approach to be determined. Therefore, internal evidence is valued as the information gathered from documentation informs clinical practice and future research directions.

Client Preference

The third component of EBP involves consideration of client preference. Clinicians deliver the best available external evidence to the client and decisions are made by supplementing that evidence with the client's preferences and values (Dollaghan, 2007). The process depends highly on the client's needs. It is the role of the clinician to develop a strong understanding of clients' preferences and give meaningful alternatives for the client's consideration and choice (Dollaghan, 2007). It is beneficial for clinicians to incorporate patients' views in their clinical decision making.

Summary of Evidence Based Practice

The above is an overview of three components of EBP in speech pathology. In the application of EBP to clinical practice all three sources of evidence are equally important. Firstly, systematic research evidence is considered to establish the effects of a treatment, thus informing clinical decision making. Traditionally, treatment choice was based on expert opinion rather than EBP (Liamputtong, 2010). However, this could reduce professional accountability. This thesis determines the best treatment for early stuttering based on an evaluation of systematic research evidence of treatment outcome reports. In speech pathology, Robey (2004) provided a useful structure in a five-phase model. Onslow, Jones, O'Brian, Menzies, and Packman (2008) further developed a method to evaluate treatment outcome reports, in particular to stuttering.

Robey's Five-Phase Model

Robey (2004) adapted a five-phase model for speech pathologists and audiologists to provide a structure for organising research evidence based on phases of a clinical trial. A clinical trial evaluates the efficacy of a treatment approach in controlled experimental conditions (Liamputtong, 2010). The five-phase model is based on the clinical outcomes research of the trial. Categorising clinical trials in speech pathology

informs health care institutions and clinicians of the stage of development of a particular approach (Dollaghan, 2007). Table 1.2 depicts Robey's five-phase model.

Essential to the phase development is the distinction between treatment efficacy and treatment effectiveness. Treatment efficacy refers to scientific evidence of the impact of a treatment approach under well-controlled experimental conditions (Dollaghan, 2007), or "Does the treatment work under ideal conditions?" The effectiveness of a treatment, in contrast, refers to evidence of the impact of a treatment as it is administered in less controlled conditions (Dollaghan, 2007), or "Does the treatment work in everyday life?" In other words, effectiveness trials are more pragmatic and relate to typical practices in comparison to efficacy trials which are controlled (Hoffman, Bennett, & Del Mar, 2010). For clinicians working in general clinical practice, the effectiveness of treatment is of interest, but efficacy must first be established in laboratory conditions. The distinction relates to the stage of development of a treatment, typically beginning in the laboratory and moving to the real world (Dollaghan, 2007).

According to Robey (2004), Phase I trials identify treatment protocols where the purpose is to detect and estimate a therapeutic effect. These studies comprise case studies, small group studies and retrospective studies. Phase I trials estimate the appropriate "dose" of the treatment and are efficacy trials. Phase II trials explore the therapeutic effect and magnitude of efficacy. In these trials, the treatment protocol is refined and consistent implementation is developed by an administration manual. The study design can be case studies or small within-group effect studies. The purpose of Phase III trials is to conduct a clinical trial for the reason of testing the efficacy of an approach. These trials are considered the gold-standard design as they include large participant numbers. Phase III studies are RCTs in which participants are randomly

allocated to experimental groups. These trials produce dependable assessments of the effect size based on pre- and post-treatment data. Phase IV research expands the applicability of the treatment protocol to include new populations and different service-delivery models. The purpose of Phase IV trials is to determine the degree of the therapeutic effect in daily clinical practice, thus testing the effectiveness of treatment. Finally, Phase V trials assess the worth of a treatment program by evaluating its cost-benefit value. By this means, health care systems can determine the population that benefits from the treatment and at what cost to the institution or participant.

Table 1.2: Robey's (2004) five-phase model and description

Phase Development	Description	Efficacy or Effectiveness
Phase I	Detect a therapeutic effect Dose of treatment Case studies	Efficacy
Phase II	Magnitude of efficacy Case studies/small participant numbers	Efficacy
Phase III	Large participant numbers RCTs	Efficacy
Phase IV	Expand treatment protocol New populations Daily clinical practice	Effectiveness
Phase V	Detect the worth or value of treatment Evaluate cost-benefit value in health research	Effectiveness

Evaluation of a Clinical Trial

Evaluating treatment evidence has become increasingly important in the field of speech pathology (Dollaghan, 2007). Clinicians continually make decisions regarding treatment plans while under the constraints of principal bodies including the government, education and financial systems (Mullen, 2007). Clinicians are therefore required to evaluate systematic research to justify treatment choices and plans. The onus stems from clinicians having to support therapeutic methods, justify continuation of

services and seek funding from health care sources (Vallino-Napoli & Reilly, 2004). However, many clinicians utilise treatments that are outdated or have not been evaluated in a systematic way (Bloodstein & Bernstein Ratner, 2008). If these practices continue, professional accountability will eventually be reduced. It is necessary, therefore, for clinicians to systematically review the literature before deciding on the treatment program to use, rather than relying on outdated treatment.

To determine the use of EBP among speech pathologists, Zipoli and Kennedy (2005) administered a questionnaire on evidence-based health care. Of the 240 respondents, only 17.7% indicated accessing research in the past 6 months. In another study, Vallino-Napoli and Reilly (2004) surveyed 697 speech pathologists and found that only 37% accessed journals daily for patient care. Findings from these studies show that only a minority of clinicians engaged in EBP. These findings are problematic, considering the role of clinicians in decision making and justification of a treatment approach. The most significant barrier for clinicians to engage in EBP was a lack of time (Vallino-Napoli & Reilly, 2004). The daily requirements of a clinician make it difficult to set aside the appropriate time required to engage with the literature. Further, the large numbers of published citations on treatment approaches are time consuming for clinicians to evaluate critically (Dollaghan, 2007). The problem, consequently, is twofold in that (a) clinicians are struggling to find time, and (b) they are unsure where to begin researching the literature. Thus it is necessary that a method that is both systematic and proficient for evaluating the literature be available for clinicians.

Onslow et al. (2008) developed a method for evaluating research evidence that reduced the burden of work for clinicians. These authors drew on recommendations from many experts in the field of stuttering research (Bloodstein, 1995; Bothe, Davidow, Bramlett, & Ingham, 2006; Conture & Guitar, 1993; Curlee, 1993; Ingham

1984; Ingham & Riley, 1998; Starkweather, 1993). They argued that the clinical trial was the “most fundamental, clinically interpretable, and useful output unit of stuttering treatment research” (Onslow et al., 2008, p. 404). In the evaluation of external evidence with respect to EBP, the levels of evidence and methodological quality are encompassed in the definition of a clinical trial by Onslow et al.

Clinical trials evaluate the efficacy of a treatment approach in controlled experimental conditions (Liamputtong, 2010). According to Onslow et al. (2008) the definition of a clinical trial must include three features. First, a clinical trial must include an entire treatment as outlined in a treatment manual or report, and therefore cannot include any modifications or be discontinued halfway. Second, a clinical trial should include outcome measures. In speech pathology at least one outcome measure must be evident, such as percentage of syllables stuttered (%SS) and/or syllables per minute (SPM). Finally, a clinical trial reduces bias. Bias is the belief that a treatment is efficacious when it is not or that a treatment is more efficacious than it is. To reduce bias, outcome measures should be evaluated by a blinded observer, a person independent of the research. Although useful for guiding clinical practice, retrospective studies which examine data from research files after the treatment is complete are not considered clinical trials because they might produce bias. Evaluating treatment outcome reports using the method of Onslow et al. is both systematic and less time consuming for clinicians. Table 1.3 describes the Onslow et al. (2008) phases of a clinical trial.

Table 1.3: Onslow et al. (2008) clinical trial phases

	Phase I	Phase II	Phase III
Ranking	Preliminary investigation Lowest level	Investigation after promising results from Phase I trials	Gold standard Highest level
Type of Treatment	New treatment	Same treatment	Same treatment
Participants	Few volunteers <10	More than >10 subjects	Relatively large, several hundreds to thousands of subjects
Trial	Case studies or small group	Can be a RCT	Is always a RCT
Aim	Primary issue “safety”	Aim to obtain a preliminary estimate of efficacy	How much better than “nothing” is this treatment?

Phases of a Clinical Trial

Onslow et al. (2008) incorporated the principles of randomisation and effect size to allocate treatments into one of three phases. In particular for stuttering treatment reports, the majority of clinical trials evidence fits within the framework of three phases rather than the five-phase model as described by Robey (2004). The phases inform the reader of the stage of development of the treatment. Onslow et al. described Phase I trials as a preliminary investigation of a new treatment. The primary goal of such trials is to determine the safety of a novel approach. Existence of a therapeutic effect is required in Phase I trials in order to proceed to the next stage of development. Participant numbers are normally below 10, as it would be unethical to deliver a novel treatment to any more subjects. Phase II trials test the safety and viability of the approach for increased subject numbers, normally greater than 10. The purpose of these trials is to establish an estimate of the number of subjects that respond to the treatment.

Replication of results is not necessary but is beneficial to estimate generalisation to a population in order to establish treatment efficacy. Phase III trials are gold-standard evidence and normally include large subject numbers, from hundreds to thousands. These trials are always RCTs comparing a treatment group to a control group, thus providing estimates of effect sizes. Phase III trials follow successful Phase II trials.

Summary

Although the cause of stuttering is unknown, researchers have developed causal theories. Theories as to the nature of stuttering have been instrumental in driving the development of treatments for clinical practice. Traditionally, choice of treatment has been based on expert opinion rather than EBP. The danger of treatment choice based on expert opinion is that it reduces professional accountability. Thus EBP has become increasingly important for clinical decision making. This thesis considers the role and application of evidence in the management of early stuttering.

Researchers agree that treating stuttering during the preschool years is important to prevent the negative consequences associated with persistent stuttering. If left untreated, stuttering can lead to mental health disorders including social phobia. Controversy exists regarding the rate of natural recovery. It is generally accepted, however, that many children recover without treatment. Factors associated with recovery including being female, a shorter time since stuttering onset, and a family history of recovery. Although these factors exist, the evidence for predictors of natural recovery is not conclusive.

Clinicians are faced with difficult decisions with regard to treating early stuttering. In that decision-making process EBP is important. Dollaghan (2007) presented a definition of EBP that encouraged equal consideration of the evidence from external, internal and client perspectives in the field of speech pathology. Researchers

benefit from the evaluation of external evidence to determine the gaps in the literature for the purpose of deriving more questions for study in order to further scientific knowledge. Clinicians also benefit from evaluation of external evidence by gaining information regarding the effects of treatments that apply directly to their clinical practice. Clinicians can then justify that practice to governing bodies. However, faced with large caseloads and vast amounts of published literature, clinicians lack the time and/or the knowledge to engage in the literature. Robey (2004) developed a system for identifying the phase of development of a treatment, establishing a method for speech pathologists to classify reports. For treatment outcome reports, Onslow et al. (2008) further developed a method of evaluating the literature encompassing both level of evidence and methodological quality in the definition of a clinical trial to determine treatment efficacy.

In the following chapter, a literature review of early stuttering treatments is conducted, highlighting reports for which clinical trials evidence is available, using the criteria of Onslow et al. (2008). Included in the evaluation is the level of evidence obtained according to the NHMRC (2009) guidelines. The purpose is to establish the best available external evidence for early stuttering treatments. This information is relevant to the choice of treatment investigated in this thesis.

CHAPTER 2

Evaluating Treatments for Early Stuttering

Over the years many intervention programs have been developed for early stuttering. The aim of the various approaches was to remediate or decrease the impact of the disorder. The approaches have greatly contributed to the body of knowledge of treatment for early stuttering and so are included in the presentation of treatments. There are different ways to classify early stuttering approaches. This thesis will classify the treatments within three distinct categories: those based on a multifactorial approach, those involving speech restructuring, and those using verbal response contingent stimulation procedures. A summary of the treatments are presented in Table 2.1.

A method for evaluating stuttering treatment reports was presented in Chapter 1 based on the definition of a clinical trial by Onslow et al. (2008). According to those researchers, a clinical trial involves a prospective study of an entire treatment program. Outcome data are collected based on at least one pre-treatment and one follow-up measure at 3 months or more when a positive outcome is reported. The outcomes involve independent speech observations beyond the clinic from recorded conversational speech.

In this chapter, a literature review of early stuttering treatments is conducted. The evidence for each report is evaluated according to the evidence contributed by clinical trials, if it exists, and the level of evidence of the report according to the NHMRC (2009) guidelines. The purpose is to establish the best available external evidence for an early stuttering treatment approach.

Multifactorial Treatments

Multifactorial theory proposes that there are multiple factors that contribute to a child's vulnerability to stuttering (Smith & Kelly, 1997). These are a combination of physiological, linguistic, psychological and environmental factors (Millard, Nicholas, & Cook, 2008). The environment is hypothesised to interact with the child's inherited or acquired neurophysiologic factors. The interaction of both can combine to stress the child in conversational and social situations. Due to these stressors, stuttering may arise or increase in such circumstances. Four treatment approach variants have been developed with a multifactorial framework, as presented below.

Group Play Therapy

Group play therapy is a treatment for early stuttering based on the assumption that stuttering children are emotionally immature, anxious and poor at self-expression (Wakaba, 1983). The aim of treatment is to improve the interpersonal relationships of stuttering children with the intention of dealing with these issues. The treatment is guided by eight principles, such that the speech pathologist must develop a friendly relationship with the child; accept the child as is; allow the child to freely express him/herself; recognise the child's feelings and reflect this back to the child in an appropriate manner; respect the child's problem solving skills; follow the child's lead; allow treatment effects to take as long as needed and make the child aware of limitations only in response to real world situations (Axline, 1947).

The current level of evidence according to the NHMRC (2009) guidelines is Level IV case studies. One study reports treatment outcomes for three children between 4 and 5 years of age (Wakaba, 1983). Post-treatment measure of stuttering frequency was available. Although clinically significant reductions in stuttering were apparent for one of the three children following intervention, the other two had a reported increase in

stuttering post-treatment. According to the Onslow et al. (2008) criteria, clinical trials evidence is not available because the current report does not meet the criteria due to the lack of follow-up outcome data.

Demands and Capacities Model

The Demands and Capacities Model (DCM) is an approach to early stuttering based on the assumption that stuttering arises when the demands on fluency exceed the child's speech motor capacity (Franken, Kielstra-Van der Schalk & Boelens, 2005; Gottwald & Starkweather, 1999). The treatment is delivered by parents, and the clinician aims to teach parents to reduce fluency demands on the child by speaking more slowly and waiting for responses. The goals of therapy are to increase the child's capacities for fluency in four domains: motoric, emotional, linguistic and cognitive.

The highest level of evidence available for this approach according to the NHMRC (2009) guidelines is an uncontrolled trial, level III-3 (Franken et al., 2005). Franken and colleagues (2005) performed a pilot study comparing 2 treatments for early stuttering including DCM and the Lidcombe Program. Altogether 12 participants were randomly assigned to the DCM group and 11 participants to the Lidcombe Program group. All children received only 12 weeks of the program. Stuttering severity and percent syllables stuttered were available both pre- and post-treatment. There were no differences reported between groups. Positive outcomes for both groups were shown after 12 weeks of treatment. According to the Onslow et al. (2008) criteria, the study is not a clinical trial because the entire treatment approach was not delivered.

Mother-Child Interaction Therapy

Mother-child interaction therapy is a treatment for stuttering that has been applied to children under six. The approach is based on the assumption that stuttering develops in response to a disruption of the relationship between the mother and child.

The clinician assists the mother to redevelop an appropriate interaction by simplifying her language, matching the child's words, correcting the child's grammar and imitating the child (Wyatt, 1969).

Wyatt (1969) reported outcomes for seven children between 2 and 5 years. Pre- and post-treatment severity ratings are available. Findings show reductions in stuttering severity for all seven children. However, no follow-up data were collected. According to the NHMRC (2009) guidelines, evidence is available in an uncontrolled trial, level III-3. According to the Onslow et al. (2008) criteria, clinical trials evidence is not available because the current report does not meet the criteria due to no reported follow-up data.

Parent-Child Interaction

The parent-child interaction (PCI) approach is a treatment program for stuttering preschool children up to 7 years of age (Millard et al., 2008), which was developed at the Michael Palin Centre in the United Kingdom. The approach facilitates parents' natural style of interaction by use of video feedback as an aid. Parents set their own targets and are guided to further develop their natural instinctive responses to help increase the child's fluency. A principle of PCI is the importance of parents' understanding of the child's needs. The child's linguistic and speech motor strengths and weaknesses are initially measured. If weaknesses are apparent in the child's planning, organising and delivering a message fluently, these aspects are explored further (Kelman & Nicholas, 2008). Another principle is that parents are already naturally supporting their child's spontaneous fluency. Through identification of fluency facilitating skills, parents become more knowledgeable about what they are already doing well to facilitate their child's fluency and how to increase those skills. Parent

involvement in the process of therapy is an essential principle to achieve reduction in the child's stuttering.

Millard and colleagues (2008) performed a study of PCI therapy. The participants were six stuttering preschool children. There were three phases of the program. Phase A was the no-treatment baseline lasting 6 weeks. Phase B was a 12-week treatment phase, 6 weeks consisting of weekly clinic-based visits and 6 weeks of home-based sessions. Phase C was a 1-year post-therapy follow-up, during which time the child and parent visited the clinic at 3-months, 6-months and finally 1-year post-therapy. Stuttering data were collected by speech recordings during the three phases. Post-treatment outcomes of percentage syllables stuttered and severity ratings were collected. After 1 year post-treatment, stuttering was reduced from a mean of 8.4%SS pre-treatment to 2.7%SS at follow-up. Four of the six children reduced their stuttering to clinically significant levels with both parents. One child reduced stuttering with only one parent and the last child did not reduce stuttering until a direct therapy program was introduced. According to the NHMRC (2009) guidelines, this study is a Level IV case study research design. The study is classified as a Phase I clinical trial according to the criteria of Onslow et al. (2008).

Millard, Edwards and Cook (2009) conducted another study of PCI therapy to explore its efficacy. The participants were six stuttering preschool children. The trial had four phases, each lasting 6 weeks. Video recordings were made while the child and parent played together at home. Each child's speech was analysed for stuttering frequency. The video recordings were made during the baseline phase (6 weeks prior to therapy), clinic-based phase (6 weeks during therapy) and home-based phase (6 weeks during therapy). The last phase was follow-up which began 3 months after the end of the home therapy and ended with a review session 6 months after the clinic phase.

Stuttering frequency measured in percentage or stuttered words were available for all recordings. Each of the six children showed a reduction in stuttering by the end of the study. According to the NHMRC (2009) guidelines, the level of evidence for this study is Level IV, a case study design for six children who received PCI treatment for stuttering. According to the Onslow and colleagues (2008) criteria, the present report is classified as a Phase I clinical trial because the subject numbers are below 10.

Speech Restructuring

Speech restructuring is a method of speaking using a novel speech pattern to facilitate fluency. Goldiamond (1965) originally described the method while using delayed auditory feedback as a contingency for stuttering. The subjects began producing a drawling speech pattern and remained stutter free when their speech was shaped to natural sounding speech. Since then, many treatment variants using speech restructuring methods have been developed, with techniques such as prolongation, rate reduction, and soft contacts. The majority of these treatments have been evaluated in research trials for adults and adolescents. However, few researchers have trialled the treatment with young children. Presented in the next section is a summary of five variants of speech restructuring that have been used with preschool-age children.

The Comprehensive Stuttering Program

Kully and Boberg (1991) developed the Comprehensive Stuttering Program at the Institute for Stuttering Treatment and Research in Canada. The program is a speech restructuring approach for stuttering that was originally developed for adolescents and adults who stutter but has also been used with children. The program components involve a programmed, prolonged speech approach incorporating techniques including easy breathing, smooth blending, light touches and gentle starts.

A study was performed on two subjects aged 4 years 8 months and 5 years 10 months (Kully & Boberg, 1991). Percent syllable stuttered measures were obtained pre- and post-treatment. Follow-up measures were available between 8 and 18 months. Both children showed clinically significant reductions in stuttering from pre-treatment to follow-up. According to the NHMRC (2009) guidelines, the highest level of evidence is Level IV, case studies. This report is not classified as a clinical trial according to the Onslow et al. (2008) criteria due to lack of independent measurement of speech recordings made beyond the clinic.

The Fluency Rules Program

The Fluency Rules Program (FRP) was originally developed for adults who stutter and later modified for children (Runyan & Runyan, 1986). The program instructs children to develop specific rules to facilitate fluency by translating anatomical and physiological concepts into child-friendly language. The rules are based on the child speaking more slowly, starting the voice gently, articulating one word at a time, reducing the length and complexity of utterances, and breathing easy.

A study using the FRP was performed with four children between the ages of 3 and 5 years. Pre- and post-treatment measures were obtained with number of stuttered words. Follow-up data were available at 12-months post-treatment for three of the four subjects. The number of stuttered words decreased for all children from pre- to post-treatment and at follow-up for three of the children. There was available evidence at Level IV case studies according to the NHMRC (2009) guidelines (Runyan & Runyan, 1986). This report is not classified as a clinical trial according to the Onslow et al. (2008) criteria because no beyond-clinic speech recordings were obtained.

The Preschool Fluency Development Program

The Preschool Fluency Development Program was developed by Culp (1984) and involves teaching children to use slow, exaggerated, easy speech with prolongation techniques. The program increases the length and complexity of utterances in a hierarchical manner while incorporating fluency distracters.

A study was performed on 14 children between the ages of 2 and 5 years (Culp, 1984). Pre- and post-treatment measures were obtained in percent disfluency. Follow-up measures were obtained 2 years post-treatment for seven of the 14 children. Group means for the 14 children showed a reduction in stuttering from pre- to post-treatment from 11% to 3% disfluency. Group means for seven children showed a reduction in stuttering from pre-treatment to follow-up from 11% to 3% disfluency. According to the NHMRC (2009) guidelines, there is evidence available at Level III-3, uncontrolled trial. According to the Onslow et al. (2008) criteria, the study is not classified as a clinical trial as no beyond-clinic measures were reported.

Intensive Stuttering Therapy Program

The Intensive Stuttering Therapy Program is a treatment approach for stuttering that has been delivered to children (Hasbrouck, Doherty, Mehlmann, Nelson, Randle et al., 1987). The program consists primarily of instructions to the client on passive airflow through the vocal cords just prior to vocalisation. Treatment is directed towards continuous airflow during speech production while concurrently identifying body tension in the speech system. To identify sources of tension in the larynx, the researchers used an electromyographic (EMG) biofeedback device to build awareness of vocal tension. The program ends with identifying individual speaking situations affecting the client's stuttering.

A study was conducted on three children aged 5 years (Hasbrouck et al., 1987). Pre- and post-treatment measures were obtained in percent stuttered words. Follow-up data are available between 6-7 months post-treatment for two of the three children. All three children reduced stuttering from pre- to immediately post-treatment under 1% stuttered words. At follow-up one of the two children regressed but not to pre-treatment severity. For this treatment approach using the NHMRC (2009) guidelines, evidence exists at Level IV case studies. According to the Onslow et al. (2008) criteria, this study is not classified as a clinical trial as beyond clinic (BC) measures were not reported.

Syllable-Timed Speech

Syllable-timed speech (STS), otherwise known as rhythmic speech training, is another treatment approach for stuttering. The approach was used to treat chronic stuttering in adults and has shown positive results with that age group (Ingham, 1984). Initially, clients were taught to produce speech in time to a beat or a metronome (Ingham, Andrews, & Winkler, 1972; Mallard, 1977). However, in later studies STS was modelled with a near normal speech rate without the use of a metronome or beat (Trajkovski, Andrews, O'Brian, Onslow & Packman, 2006; Trajkovski, Andrews, Onslow, Packman, O'Brian, et al., 2009).

Trajkovski et al. (2006) reported using STS to treat stuttering in a 3-year-old boy in a non-programmed parent training approach. The child's speech was recorded just prior to the start of treatment and 5 and 10 weeks into the treatment phase of the program. Post-treatment outcomes were measured in percentage syllable stuttered and severity ratings. At the 16th week of treatment the child reached 0%SS within the clinic, a clinically significant value. The level of evidence is Level IV, a case study according to the NHMRC (2009) guidelines. According to the Onslow et al. (2008) criteria of a clinical trial, this study is classified as Phase I.

Another study of STS was conducted by Trajkovski et al. (2009) with three preschool children. The protocol was a multiple baseline design for three participants. Post-treatment outcomes were measured in percentage syllable stuttered. Stage 1 entailed reducing stuttering frequency to below 1.0%SS for 3 weeks before progressing to Stage 2, which was the gradual withdrawal of STS practice. At the end of Stage 1, all children reduced the percentage of stuttering to clinically significant levels of below 1.0%SS in a mean of 8.6 clinic visits. Both of the reported Phase I clinical trials on STS are early prospective studies. The level of evidence according to the NHMRC (2009) guidelines is Level IV, case studies. According to the Onslow et al. (2008) criteria, this study is classified as a Phase I trial.

A Phase II clinical trial of STS was conducted by Trajkovski et al. (2011) with 17 preschool aged children. The program was implemented as indicated above. Progression to Stage 2 was based on %SS measures of less than 1.0%SS over two consecutive fortnightly visits and average weekly SRs less than 2.0%SS. The primary outcome measure was %SS in beyond-clinic audio recordings occurring at pre-treatment, entry to Stage 2, 6-months post Stage 2, and 12-months post Stage 2. Altogether eight children completed Stage 1 of the program and nine children dropped out before completing Stage 1. For the eight children who completed, the mean %SS reported was 6.0 pre-treatment, 1.3 at entry to Stage 2 and 0.2 at 12-months follow up. The mean number of clinic visits to complete Stage 1 was 12.4. The mean number of clinic hours was 8.0. The level of evidence according to NHMRC (2009) guidelines is level III-3, an uncontrolled trial, as there was no comparison to a control group.

Verbal Response Contingent Stimulation

Verbal response contingent stimulation (VRCS) is a behavioural treatment model that requires use of verbal feedback contingent on stutter-free or stuttered speech.

Early researchers suggested that punishment was related to the development of stuttering in preschool children (Bloodstein, 1969; Van Riper, 1971). Therefore, the traditional emphasis in therapy was placed on counselling parents to reduce feedback contingent on stuttering (Reed & Godden, 1977). However, a study reported a reduction in stuttering after the presentation of punishment (Martin, Kuhl, & Haroldson, 1972). Further studies of stuttering recovery suggested that the cessation of stuttering was due to direct feedback including asking the client to “slow down” (Shearer & Williams, 1965; Sheehan & Martyn, 1966).

VRCS is based on operant principles of stuttering. These principles are derived from an extensive body of knowledge (Bloodstein & Bernstein Ratner, 2008) that uses operant responses contingent on stutter-free or stuttered speech. The first experiment that used operant methods on stuttering behaviours was performed by Flanagan, Goldiamond and Azrin (1958), who demonstrated the effects of response contingent stimulation on three male subjects who stuttered, aged 15, 22 and 37 years. In the first part of the study, the researchers presented a 1-second, 105 dB tone contingent on stuttering and measured the effects. In the second part of the study, they played a continuous tone and removed the tone for 5 seconds when the subject displayed stutter-free speech. When the tone was applied, stuttering rates increased. However when the tone was removed, stuttering rates decreased to escape the punishing sound. Subsequent to this study, further research expanded the use of VRCS procedures on stuttering behaviours.

Early VRCS Treatments

Martin et al. (1972) conducted an experiment using puppets with two preschool children. A puppet was mounted in an illuminated box and after a series of conversations the puppet disappeared when the child stuttered. Stuttering reductions for

both children were immediate following this time-out procedure. The researchers conversed with the children in the clinic during monthly probe sessions. Stuttering severity was also measured in the home environment beyond the clinic. Outcomes were measured in number of words stuttered. For both children, the number of words stuttered was greatly reduced at home and in the probe sessions. The reduction in stuttering was maintained for almost a year after treatment. The level of evidence is Level IV, a case study according to the NHMRC (2009) guidelines. According to the Onslow et al. (2008) criteria, this report is classified as a Phase I clinical trial.

Reed and Godden (1977) performed a multiple baseline experiment with two preschool children, who were seen for individual treatment sessions for 20 minutes each week. The children conversed with the researchers in spontaneous conversation. The researchers delivered the phrase “slow down” contingent on stuttering. Outcomes were measured in number of words stuttered. Stuttering decreased and remained below pre-treatment levels for the remainder of the treatment session for both children. The reduction of stuttering generalised outside of the clinic environment. According to the NHMRC (2009) guidelines, this study is classified as a Level IV, case study. According to the Onslow et al. (2008) criteria, this study is classified as a Phase I clinical trial.

The Lidcombe Program of Early Stuttering Intervention

The majority of clinical trials evidence available comes from research on the Lidcombe Program, a model of VRCS procedures for early stuttering (Onslow, Packman & Harrison, 2003). Contingencies for stutter-free and stuttered speech are delivered by parents in the form of verbal feedback, resulting in the alleviation of stuttering. Full details of the Lidcombe Program are presented in Chapter 3. According to the Onslow et al. (2008) criteria, there are three Phase I studies (Harrison, Wilson & Onslow, 1999; Onslow, Costa & Rue, 1990; Wilson, Onslow & Lincoln, 2004)

establishing the viability of the program with pre-school children. Phase II evidence is available from four research reports (Lewis, Packman, Onslow, Simpson & Jones, 2008; Miller & Guitar, 2009; Onslow, Andrews & Lincoln, 1994; Rousseau, Packman, Onslow, Harrison & Jones, 2007). There are two Phase III RCTs of the Lidcombe Program (Jones, Onslow, Packman, O'Brian, Hearne, et al., 2008; Jones, Onslow, Packman, Williams, Ormond, et al., 2005).

Phase I Clinical Trials

Onslow et al. (1990) published the first Phase I clinical trial of the Lidcombe Program. The level of evidence according to the NHMRC (2009) guidelines is Level IV case studies, with four preschool children. The purpose of the study was to report within- and beyond-clinic speech data from children who received the treatment. Audiotape recordings were gathered both within and beyond the clinic, to assess generalisation of fluency in the daily environment. Speech outcome data were measured for %SS and SPM. All four subjects achieved reductions in stuttering greater than that reported for adult treatment programs, in fewer clinical hours. The program's effects proved beneficial for the small number of children in this study. The preliminary investigation sparked greater interest in larger group studies.

The Lidcombe Program has been adapted using telehealth procedures for a child unable to attend a speech clinic. A Phase I clinical trial of the Lidcombe Program was undertaken using telephone contact with a family that was isolated from clinical treatment services (Harrison et al., 1999). The subject was a boy aged 5 years 10 months who had been stuttering severely for 4 years. The treatment occurred by telephone contact between the treating clinician and the family. The family mailed audio and videotape recordings of the boy's speech to the clinician for analysis. Speech outcome data were obtained by %SS and SPM measures. After 25 phone consultations over a

period of 9 months, the boy reached near-zero levels of stuttering. He maintained these clinically significant levels for 23 months post-treatment. The success of distance therapy for this child was encouraging as it provided preliminary evidence for an alternative service delivery approach for families unable to access a speech clinic. The level of evidence was Level IV case study, according to the NHMRC (2009) guidelines.

In another Phase I experiment conducted by Wilson et al. (2004), five preschool-aged children were treated for stuttering using telehealth procedures. Unlike the previous Phase I study, video training material was provided to the parents. Speech outcome measures were collected at 1 week, 1 month, 2 months, 4 months, 8 months, 12 months and 13 months post-treatment. Speech outcome data were measured for %SS and SPM. The number of sessions required to complete Stage 1 were reported between 3 and 34 consultations. Four of the five children required consultations that exceeded established benchmarks for standard within-clinic treatment (Jones, Onslow, Harrison, & Packman, 2000). The findings suggested that telephone consultations might be less efficient than standard face-to-face treatment. However, the results showed that treatment in the telehealth format could be a viable option for families unable to access standard services for stuttering treatment. The level of evidence provided by this study was Level IV case study, according to the NHMRC (2009) guidelines.

Phase II Clinical Trials

In 1994, Onslow et al. performed a Phase II trial of the Lidcombe Program, after preliminary Phase I data (Onslow et al., 1990) had shown encouraging results. Assignment to experimental or control groups was determined in a non-random way, by clinic site location. The responses of a group of 12 preschool children who stuttered were compared to those of 11 participants in a control group. The treatment group received the entire Lidcombe Program treatment until completion of Stage 2. Outcome

measures were based on beyond-clinic audio-taped recordings. Both %SS and SPM data were obtained from the beyond-clinic tapes. Findings showed that the children completed Stage 1 in a median of 10.5 1-hour clinic visits. At the end of Stage 2, the 12 children achieved median %SS scores below 1.0. The findings were encouraging and suggested that the program might be efficacious for young children who stutter. Further, the trial concluded that the Lidcombe Program might be a cost-efficient option for health care professionals treating early stuttering. The level of evidence according to the NHMRC (2009) guidelines was Level III-2, non-randomised trial.

Rousseau et al. (2007) performed a Phase II clinical trial of the Lidcombe Program to measure predictors of treatment time during Stage 1 of the program. Altogether 29 children completed Stage 1 of the program, with a more rigorous criterion to end Stage 1 of near-zero stuttering for three consecutive clinic visits. Previous to this study that criterion had not been stipulated. Outcome measures of %SS were collected on four occasions; pre-treatment and 6, 12 and 24 months after the start of Stage 2. After completion of Stage 1, a large treatment effect was revealed for all children. Findings showed that phonological development did not predict treatment time. However, a higher receptive language score was associated with longer treatment time and a higher MLU was associated with a shorter treatment time. The median treatment time to complete Stage 1 was 16 clinic visits, which was higher than previous experimental studies when the median time was found to be 11 clinic visits (Jones et al., 2000; Kingston, Huber, Onslow, Jones, & Packman, 2003). The reason for the higher value was suggested to be the more rigorous criteria for ending Stage 1. The level of evidence according to the NHMRC (2009) guidelines was Level III-3 an uncontrolled trial, as there was no comparison to a control group.

A Phase II distance therapy clinical trial of Lidcombe Program was performed by Lewis et al. (2008). The aims were to evaluate the efficacy of telehealth delivery of the Lidcombe Program compared to a control group. In total 22 children were randomised, 9 to the experimental group and 13 to the no-treatment control group. The primary outcome measure was %SS based on audio speech samples beyond the clinic. The children in the treatment group showed a 69% greater decrease of stuttering frequency at 9 months post-randomisation compared to the control group. The findings were consistent with the two previous studies of the benefits of telehealth delivery. Although the telehealth format required more clinic visits than standard clinic treatment, it provided families from remote areas without access to speech clinics an option for receiving treatment services. The level of evidence according to the NHMRC (2009) guidelines was Level II, RCT.

A Phase II clinical trial was performed by Miller and Guitar (2009). The purpose was to determine long-term outcomes of 15 children treated with the Lidcombe Program. The clinicians were inexperienced with the Lidcombe Program and independent of the developers of the program. The children were assessed for %SS prior to treatment and at follow-up periods ranging from 12 to 58 months following the completion of Stage 1. Outcome measures of %SS both within and beyond the clinic were collected by audio-tape recordings. Of the 15 children, 11 were not stuttering at long-term follow-up and the remaining 4 had mild or very mild stuttering. The median treatment time to complete Stage 1 of the program was 17 clinic visits. The results were favourable for performing larger scale evaluations of the program's effects to verify the validity of the results. The level of evidence according to the NHMRC (2009) guidelines was Level III-3 an uncontrolled trial, as there was no comparison to a control group.

Phase III Clinical Trials

A Phase III RCT of the Lidcombe Program was conducted by Jones et al. (2005). The purpose was to evaluate the efficacy of the Lidcombe Program by comparison to a control group. The control group received no treatment. The participants were 54 preschool children who were prospectively randomised into either the treatment or control group. Altogether 29 were in the treatment arm and 25 in the control arm. There were two drop-outs from the treatment arm and five from the control arm. Children in the treatment arm were given the entire Lidcombe Program (Stages 1 and 2). Outcome measures were %SS in audio-taped beyond-clinic speech samples. At 9 months post-randomisation, children in the treatment arm had 1.5%SS compared to the control arm of 3.9%SS, giving an effect size more than double the minimal worthwhile difference as stipulated in the protocol. These results provided evidence that the Lidcombe Program was an efficacious treatment for early stuttering. The level of evidence according to the NHMRC (2009) guidelines was Level II RCT.

An extended follow up of the previous Phase III RCT was reported by Jones et al. (2008). The purpose was to measure the long-term effects of the Lidcombe Program. Of the original 29 children in the treatment arm, 20 participated in the extended follow-up. Within the cohort of 20 children, 16 maintained near-zero levels of stuttering at a mean of 5 years post-randomisation. Four children did not maintain low levels (<1.0%SS) of stuttering and were classified as having relapsed. Reasons for relapse were suggested to be associated with the removal of parental verbal contingencies during signs of increased stuttering. The findings were that the long-term success rate for the children in this study with the Lidcombe Program was 86%. Further, it was found that most of these children were able to complete the Lidcombe Program and

maintain near-zero levels of stuttering for a long period of time. The level of evidence according to the NHMRC (2009) guidelines was Level II, RCT.

Summary of Early Intervention Approaches

The review of early stuttering treatment reports presented here has indicated both the level of evidence according to the NHMRC (2009) guidelines and the available clinical trials evidence based on the criteria of Onslow et al. (2008). The multifactorial model has two available Phase I clinical trial reports for PCI treatment. In speech restructuring, there are three available clinical trials evidence for STS. Finally, for VRCS procedures, there are 11 available clinical trial reports, two for early VRCS procedure reports, and nine for the Lidcombe Program. Of these, three are classified as Phase I, four as Phase II and two as Phase III clinical trials. Table 2.1 provides a view of early stuttering treatment reports, the current level of evidence, and whether clinical trials evidence exists.

Table 2.1. Multi-factorial, speech restructuring, and verbal response-contingent stimulation models of early stuttering treatment and current level of evidence.

Treatment	Author/date	NHMRC (2009) Level of Evidence	Clinical Trial Classification/Phase	Type of Approach
Group play therapy	Wakaba (1983)	Level IV Case study	None due to lack of follow-up outcomes	Multifactorial
The Demands and Capacities Model	Franken et al. (2005)	Level III-3 Uncontrolled trial	None as the entire treatment approach was not delivered	Multifactorial
Mother-child interaction therapy	Wyatt (1969)	Level III-3 Uncontrolled trial	None due to lack of follow-up outcomes	Multifactorial
Parent-child interaction therapy	Millard et al. (2008)	Level IV Case study	Phase I	Multifactorial
Parent-child interaction therapy	Millard et al. (2009)	Level IV Case study	Phase I	Multifactorial
The Comprehensive Stuttering Program	Kully and Boberg (1991)	Level IV Case study	None due to lack of beyond clinic data	Speech restructuring
The Fluency Rules Program	Runyan and Runyan (1986)	Level IV Case study	None due to lack of beyond clinic data	Speech restructuring

The Preschool Fluency Development Program	Culp (1984)	Level III-3 Uncontrolled trial	None due to lack of beyond clinic data	Speech restructuring
Intensive Stuttering Treatment Program	Hasbrouck et al. (1987)	Level IV Case study	None due to lack of beyond clinic data	Speech restructuring
Syllable-timed speech	Trajkovski et al. (2006)	Level IV Case study	Phase I	Speech restructuring
Syllable-timed speech	Trajkovski et al. (2009)	Level IV Case study	Phase I	Speech restructuring
Syllable-timed speech	Trajkovski et al. (2011)	Level III-3 Uncontrolled trial	Phase II	Speech restructuring
Puppet experiment	Martin et al. (1972)	Level IV Case study	Phase I	VRCS
Response-contingent stimulation	Reed and Godden (1977)	Level IV Case study	Phase I	VRCS
Lidcombe Program	Onslow et al. (1990)	Level IV Case study	Phase I	VRCS
Lidcombe Program	Harrison et al. (1999)	Level IV Case study	Phase I	VRCS
Lidcombe Program	Wilson et al. (2004)	Level IV Case study	Phase I	VRCS
Lidcombe Program	Onslow et al. (1994)	Level III-2 Non-randomised trial	Phase II	VRCS
Lidcombe Program	Rousseau et al. (2007)	Level III-3 Uncontrolled trial	Phase II	VRCS
Lidcombe Program	Lewis et al. (2008)	Level II Randomised controlled trial	Phase II	VRCS
Lidcombe Program	Miller and Guitar (2009)	Level III-3 Uncontrolled trial	Phase II	VRCS
Lidcombe Program	Jones et al. (2005)	Level II Randomised controlled trial	Phase III	VRCS
Lidcombe Program	Jones et al. (2008)	Level II Randomised controlled trial	Phase III	VRCS

Summary

This chapter presented a literature review of treatment options for early stuttering under the categories multifactorial, speech restructuring and VRCS procedures. Included in the presentation of treatments was a description of the current level of evidence according to the NHMRC (2009) guidelines. Further, clinical trials evidence was provided based on the methods described by Onslow et al. (2008). As indicated in Chapter 1, many clinicians have concerns over the choice of treatment program for early stuttering. With regard to systematic evidence from an EBP approach, the highest level of evidence was seen to relate to VRCS procedures. Further, clinical trials evidence was more abundant for treatments in this category than for multifactorial or speech restructuring treatments. Of all treatment approaches, evidence for the Lidcombe Program showed both the highest level of evidence according to the NHMRC (2009) guidelines and clinical trials evidence according to the Onslow et al. (2008) criteria. Therefore, the treatment choice in this thesis for the empirical studies presented in Chapters 4 and 5 was the Lidcombe Program. The following chapter explores treatment process research of the Lidcombe Program. The purpose is to provide greater detail relevant to the evidence base for this treatment approach.

Chapter 3

The Lidcombe Program of Early Stuttering Intervention

The Lidcombe Program is a direct intervention for early stuttering. The program was developed and manualised¹ (Packman, Onslow, Webber, Harrison, Lees, Bridgeman, et al., 2010) by researchers in Australia and is widely used by clinicians around the world (Onslow et al., 2003). The approach is a behavioural treatment for stuttering children under the age of 6. The program is implemented by parents who are guided by the clinician in the delivery of treatment goals. Parents learn to use verbal contingencies for stutter-free and stuttered speech. These contingencies are delivered to the child in structured and unstructured treatment activities in the home environment.

This chapter outlines the rationale, main components and goals of the Lidcombe Program, providing a detailed account of the treatment procedures adopted in the empirical research presented in this thesis. This chapter also provides an account of the available evidence base for this approach to intervention across all aspects of EBP.

Rationale

The Lidcombe Program does not assume a perspective on the cause, nature or progression of stuttering (Onslow et al., 2003). However, the rationale of the program is based on extensive literature on operant methods influencing stuttered speech (Bloodstein & Bernstein Ratner, 2008). Operant methods utilise a stimulus, such as verbal or nonverbal feedback, contingent on behaviour. In the Lidcombe Program parents provide the child with five verbal contingencies for stuttered or stutter-free utterances. The child increases fluent speaking and reduces stuttering behaviours until stuttering reduces to insignificant levels.

¹ The most recent version of the manual has been updated and published in 2011. This thesis employed methods from the 2010 manual.

Principles

The Lidcombe Program is a treatment implemented by parents, who carry out all activities at home on a daily basis. The role of the clinician is to mentor the parent and provide feedback in the effective management of the child's stuttering. Each day, parents measure the child's severity of stuttering with home severity ratings (SRs) on a scale of 1-10, where 1=no stuttering, 2=extremely mild stuttering and 10=extremely severe stuttering. The clinician is guided by the home measurements and makes decisions about the direction and movement of treatment goals based on these ratings. Another principle is that children must enjoy the treatment process. Children are not asked to modify their speaking rate or create goals during the therapeutic process. They merely must participate and in doing so, have fun. If the child is not having fun, rapid changes to the program must be employed to ensure the child stays on target with the program principles.

Lidcombe Program Components

Parental Verbal Contingencies

There are five verbal contingencies in the Lidcombe Program that are applied contingent on the child's response for stutter-free speech or unambiguous stuttering. Stutter-free speech can be acknowledged by saying, "that was smooth", or praised by saying, "great, no bumpy talking" or requested to be self-evaluated by saying, "was that smooth?" Unambiguous stuttering can be acknowledged by saying, "that was a bit bumpy" or requested to be self-corrected, by saying, "oops, can you say that again smoothly?" To ensure that the Lidcombe Program remains positive for the child, most verbal contingencies are for the child's stutter-free speech.

Measurement

The two primary measurements in the Lidcombe Program are objective stuttering frequency and subjective severity ratings. Stuttering frequency is measured by the clinician in percentage of syllables stuttered (%SS). The clinician uses a two-button press device or another counting mechanism to determine the numbers of syllables and stutters while the child converses for approximately 10-minutes of speech. The clinician calculates the percentage of syllables stuttered (%SS) at each clinic visit and records the weekly value for comparison to previously obtained measures.

Severity ratings (SRs) are perceptual measurements of stuttering that are rated on a scale of 1-10, where 1=no stuttering, 2=extremely mild stuttering and 10=extremely severe stuttering. Parents rate the severity of the child's stuttering on a daily basis in the natural home environment and record them on a severity rating chart. The parental severity ratings are used for discussion regarding treatment progress of the child.

Weekly Clinic Visits

The program manual recommends weekly clinic visits during Stage 1 of the program (Packman et al., 2010). The parent and child visit a clinician each week for treatment between 45 minutes and 1 hour.

Treatment in Structured and Unstructured Conversations

In structured treatment conversations, parents play a sit-down game with the child for a short period of time. The parent chooses an appropriate game and engages with the child in order to maximise stutter-free speech. The structured conversations occur daily and sometimes concurrently with unstructured conversations. During unstructured conversations the parent delivers verbal contingencies in the child's

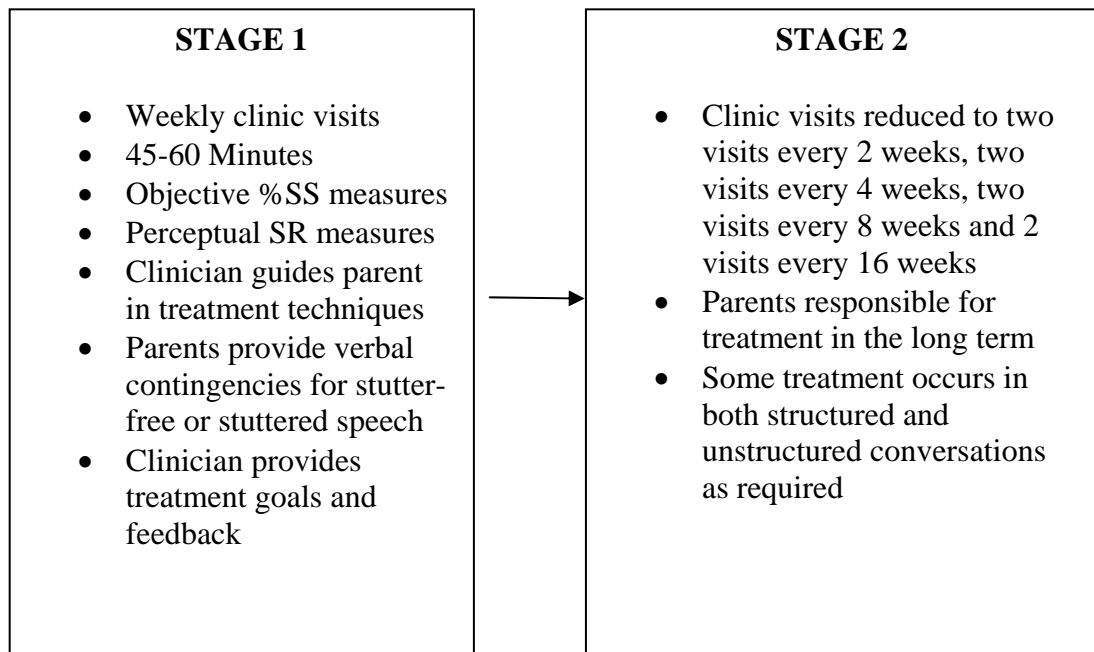
everyday life speaking situations. The goal of unstructured conversations is to generalise stutter-free speaking to all speech environments.

Primary Goals

There are two Stages of the Lidcombe Program. In Stage 1, the child and parent visit the clinic once a week until the child reaches near-zero stuttering. The criteria for ending Stage 1 according to the manual are “(1) %SS less than 1.0 within the clinic, and (2) Severity rating (SR) scores for the previous week of 1 or 2, with at least four of these being 1” (Packman et al., 2010, p. 9). These criteria need to be achieved for three consecutive clinic visits. Once the child has passed both criteria, Stage 2 of the program begins.

In Stage 2, the goal is for the child to maintain the above criteria for a long period of time. In Stage 2, clinic visits are decreased systematically. The first two visits are scheduled 2 weeks apart, the following two visits are 4 weeks apart, the following two visits are 8 weeks apart and the last two visits are 16 weeks apart, for a total of 1 year after the completion of regular Stage 1 visits. If at any Stage 2 visit the child does not maintain near-zero stuttering, the clinician may see the child earlier than the regularly scheduled Stage 2 visit. In a file audit of 25 children who completed Stage 2 of the Lidcombe Program it was found that 4 children returned to Stage 1 as recommended by the clinician (Onslow et al., 2003). The findings showed that it was rare for children to return to weekly Stage 1 clinic visits. Figure 3.1 presents the different procedures adopted in Stage 1 and Stage 2 clinic visits (Onslow et al., 2003).

Figure 3.1: Progression from Stage 1 to Stage 2 visits, adapted from Onslow et al. (2003)



Treatment Process Research

Many clinicians around the world use the Lidcombe Program to treat early stuttering. Given the high rate of natural recovery, debate exists regarding when to treat early stuttering. Clinical trials evidence of the Lidcombe Program was provided in Chapter 2, along with the current levels of evidence according to the NHMRC (2009) guidelines. The following review is of research into the Lidcombe Program that investigates evidence related to natural recovery, long-term impact, psychological impact, social validity, effects on speech and language, cultural impact, parent perception, school-age adaptations and predictors for treatment recovery.

Natural Recovery

Harris, Onslow, Packman, Harrison, and Menzies (2002) investigated whether the effects of the Lidcombe Program were better than those of natural recovery. The participants were 23 children who were randomised into one of two groups, either control or treatment. Children were compared over 12 clinic visits, approximately the

mean number of clinic visits to complete Stage 1 (Jones et al., 2000). All subjects were required to record three beyond-clinic audio-recordings and one within-clinic video recording. Children in the control group returned 12 weeks post-randomisation. Results showed that children in the treatment group reduced stuttering twice as much as the control group, at a statistically significant level ($p < 0.001$). Therefore, the Lidcombe Program was demonstrated to have had a positive impact on the frequency of stuttering for children in the treatment group which exceeded that of natural recovery.

Long-Term Impact

A long-term outcome study was performed by Lincoln and Onslow (1997). Children from the preliminary Phase I (Onslow et al., 1990) and Phase II (Onslow et al., 1994) studies were assessed 2-7 years post-treatment. Altogether 43 participants between 2 and 5 years who had been treated for stuttering were included in this research. The purpose was to measure the long-term outcomes of the Lidcombe Program. The parents were required to make three 10-minute recordings within the span of a month. Results showed that near-zero stuttering levels were achieved and maintained for the participants up to 7 years after the initial treatment.

Psychological Impact

The Lidcombe Program is a direct approach for early stuttering that requires a request for correction of stutters from the child from time to time. Therefore, concerns have been raised that the program might have adverse psychological effects on a child. For example, verbal contingences may affect the child's self-esteem (Stewart, 1996) and might cause the child to think "I must not stutter" (Cook & Rustin, 1997, p. 255). Woods, Shearsby, Onslow, and Burnham (2002) investigated the psychological impact of the Lidcombe Program, to identify any evidence of adverse psychological effects on children who had received the program. The subjects were eight preschool children who

were treated successfully with the program. The Child Behaviour Checklist (CBCL) was given to parents post-treatment as a measurement tool used to detect anxiety, aggression, withdrawal or depression from the child. Another checklist given to parents during the course of treatment was the Attachment Q-Set (AQS) which measured the changes in the quality of the parent–child attachment relationship. The CBCL findings showed no systematic post-treatment negative effects in the child. Rather, the findings suggested an improvement in scores after treatment. The AQS data showed no changes in the quality of the attachment relationship between the child and parent after treatment. The results provided no indication that the Lidcombe Program was psychologically harmful for children.

Social Validity

Lincoln, Onslow and Reed (1997) evaluated post-treatment speech samples of children who had received the Lidcombe Program. The purpose was to measure the impact of change or social validity in the speech of children who had completed Stage 1 and 2 of the program. Clinicians experienced in stuttering and unsophisticated adult listeners participated in this study. The first part of the study compared post-treatment %SS for pre-school and school-age children with non-stuttering control children. The second part compared the number of stuttering and fluent moments in both groups. Findings showed that %SS measures between the groups were not significantly different. In fact, the children in the control group were judged as having more stuttering than the children who had completed the Lidcombe Program. One possible reason for this is because experienced clinicians may be more sensitive to stuttering due to the nature of their work in the field. The children who had completed the program were not judged as “stuttering” by unsophisticated listeners. The treatment outcomes were judged to be socially valid.

Child and Parent Speech and Language

In the Lidcombe Program, parents are not instructed to change their speech rate while conversing with the child. However, concerns have been raised that the positive treatment effects from the program might be due to changes in parent and child speech and language. This possibility was investigated by Bonelli, Dixon, and Bernstein Ratner (2000), who measured child and parent speech and language before and after delivery of the program. Conversational speech samples of nine parents and nine children were evaluated both pre- and post-treatment. Pre-treatment conversations were recorded 1 week to 1 month before treatment and post-treatment conversations recorded 1 week to 1 month after the completion of Stage 2. The findings indicated that parental speech rate increased in post-treatment samples. The language measures collected were mean length of utterance, development sentence score, number of different words and requests for clarification or information. All children were found to be within or above developmental expectancies. This study did not support the suggestion that treatment induced changes in the language functioning of the parent or child.

Cultural Impact

Latterman, Euler and Neumann (2008) in Germany evaluated the impact of the Lidcombe Program on German-speaking preschool children. Forty-five children were randomly allocated into either a treatment (22 children) or no-treatment (23 children) group. Speech samples were recorded pre-treatment and 16 weeks later for children in both groups. Findings showed that children in the treatment group had significantly higher decreases in %SS compared to the no-treatment group. At the end of 16 weeks, only three children who received the Lidcombe Program reached Stage 2. Children who received treatment had a mean within-clinic (WC) reduction of 6.8%SS compared to

1.6%SS in the no-treatment group. The authors concluded that the short-term impact of the Lidcombe Program for stuttering German preschool children was beneficial.

In a study in Saudi Arabia, Rifaie, Hasan, Saber and Kaddah (2009) evaluated the effects of the Lidcombe Program on stuttering preschool children. The treatment group subjects were 10 children between the ages of 4 and 7 years. The control group subjects were 10 children matched for gender, age and stuttering severity. The Lidcombe Program was delivered for a period of 25 weeks. Speech outcome data was measured in percent stuttered words and severity ratings. After 25 weeks, the children in the Lidcombe Program group reduced stuttering from 27.8% to 2.1% stuttered words, a clinically significant reduction ($p < 0.05$). The control group showed a non significant ($p > 0.05$) reduction in stuttering after 25 weeks. The authors concluded that the Lidcombe Program was an effective treatment for Saudi stuttering children.

Implementation with School-Age Children

Lincoln, Onslow, Lewis, and Wilson (1996) undertook a study of the Lidcombe Program on school-age children. Previously, treatment for this age group had been influenced by adult programs but no simple operant approach had been explored. The treatment was the Lidcombe Program as described by Onslow et al. (1994). The purpose of this study was to investigate the effect of a direct operant therapy approach on school-age children. Eleven children between the ages of 7 and 12 participated in this study. Speech assessments were conducted in a series of pre- and post-treatment audio-recordings. Pre-treatment recordings occurred 2 months, 1 month and 1 week prior to treatment. Post-treatment audio-recordings occurred 1 week, 1 month, 2 months, 3 months, 4 months, 6 months, 9 months and 12 months after completion of Stage 1. The median number of clinic visits to complete Stage 1 of the program was 12, far less than in previous treatments for school-age children. All children maintained reduced

stuttering rates at 12 months post-Stage 1. However, five of the 11 children failed to meet Stage 2 criteria at some time post-treatment. Findings showed that the Lidcombe Program might be effective for some children within this age group.

Koushik, Shenker, and Onslow (2009) conducted a follow-up study of the Lidcombe Program for school-age children. The purpose was to establish the viability of the program and to determine how many clinic visits were required to significantly reduce stuttering frequency. The school-age children had received the Lidcombe Program but some adaptations had been made in cognisance of their advanced age. For example, verbal contingencies were delivered by self-reinforcement rather than parent delivered. The participants were 11 school-age children between 6 and 10 years of age who were treated with the Lidcombe Program. Pre-treatment audio-recorded samples were obtained within the clinic 1-2 weeks prior to treatment. Follow-up assessments were conducted in random telephone calls to the children in their home environment 9-187 weeks post Stage 1. Stuttering frequency was reduced from a mean of 9.2%SS pre-treatment to 1.9%SS at follow-up. The results were obtained in a median of eight clinic visits (range 6-10 visits). Findings for these children showed that significantly fewer Stage 1 clinic visits were required than for preschool children.

Parent Perception

The Lidcombe Program is a parent training program for early stuttering and thus the program components cannot be executed effectively without the parent. The role of the clinician is to act as a guide aiding the parent in the delivery of the program. Since the burden of work falls on the parent for effective delivery of treatment, research into parents' perspectives of the program is highly valued. The following studies involved interviewing parents regarding their perception of the Lidcombe Program.

Hayhow (2009) interviewed the parents of 14 stuttering children using a qualitative methodology. The parents were interviewed one to two times at different stages of the Lidcombe Program to assess whether their perceptions changed throughout the treatment process. In the analyses, three themes emerged from the interviews: 1) the program as a straightforward journey, 2) the program starting well but hitting problems, and 3) the program as a problematic journey from the start. The majority of parents indicated that the program was either straightforward from the start or began well and faced problems along the way. One parent indicated that the service delivery of weekly clinic visits became burdensome over a longer period of time. Only two parents found the treatment process problematic from the start. The findings from this study were among the first to provide insight into parents' experience with the Lidcombe Program.

Goodhue, Onslow, Quine, O'Brian, and Hearne (2010) performed a phenomenological study exploring the perception of the Lidcombe Program of 16 mothers. The mothers were interviewed pre-treatment and then throughout a 6-month time frame during the treatment process. The interviews were conducted face to face and by telephone. The findings were organized into themes: 1) treatment implementation, 2) perception of the program, and 3) emotions reported. Overall, the interviews revealed that the mothers perceived the program to be effective, theoretically easy and requiring a commitment for the program to work. The children generally enjoyed the therapy and showed an increase in confidence. However, some negative emotions were expressed, including parental anxiety and distress, especially with increased severity of stuttering. Solutions to address the problems were provided by the clinicians to all mothers. The study presented findings helpful for clinicians in the guidance of parents using the Lidcombe Program.

Predictors of Treatment Time

There are two large group retrospective studies examining the effects of the Lidcombe Program in clinical communities. The first study was conducted in Australia by Jones et al. (2000) and the second in the United Kingdom by Kingston et al. (2003). The purpose of these studies was to determine if the duration of treatment with the Lidcombe Program could be predicted by examining case variables from clinical files. The data from these studies were combined and meta-analysis performed.

Australian Study

Jones et al. (2000) examined the case files of 250 preschool children who had successfully completed Stage 1 of the Lidcombe Program. The aim was to determine whether there was a relationship between predictor variables and treatment time to Stage 2. The predictor variables examined were gender, age at first treatment session, time between stuttering onset and first treatment session (onset-to-treatment interval) and %SS at first treatment session. Statistical analyses were performed for all data variables for the 250 case files unless data were missing. The median duration of clinic visits, where 50% of the children completed Stage 1, was 11 sessions. Pre-treatment severity, measured by %SS, was found to be a significant predictor of treatment time. Children with more severe stuttering (5.0%SS or more) required more clinic visits to reach Stage 2 than children with less severe stuttering (<5.0%SS). A non-significant predictor was the onset-to-treatment interval. Findings indicated that a longer period between onset and treatment might be associated with shorter duration of treatment. This was contrary to the findings of Starkweather and Gottwald (1993) who reported a relationship between longer onset-to-treatment intervals and increased duration of treatment. However, in both studies increased severity of stuttering predicted longer treatment times.

United Kingdom Study

A replication of the Jones et al. (2000) study was performed by Kingston et al. (2003) in the United Kingdom. The purpose was to determine whether the program's effects could be reproduced in a country other than where the program was developed. Further, direct replication enabled pooling of the Australian and British cohorts in order to perform meta-analysis. The case files of 66 children who had completed Stage 1 of the Lidcombe Program were examined for the same variables: gender, age at first treatment session, onset-to-treatment interval and %SS at first treatment session. Kingston et al. used a similar methodology to that used by Jones et al. The median duration of treatment visits was 11, which was identical to the original study. Further, the severity of stuttering at first treatment visit was found to be a significant predictor of treatment time. Children with a higher pre-treatment severity took longer to complete Stage 1 than those with lower pre-treatment severity of stuttering.

Meta-analysis

Kingston et al. (2003) pooled the data from the Australian and British cohorts (N=316) for meta-analysis. Meta-analysis increased the sample size and thus the statistical power of the reported outcomes. The meta-analysis showed that children whose pre-treatment stuttering was more severe (5.0%SS or more) were 3.5 times as likely ($p<0.0001$) to require more clinic visits to complete Stage 1 than children whose pre-treatment stuttering was less severe. Further, children who had been stuttering for less than 12 months had twice the odds ($p=0.013$) of requiring more clinic visits to complete Stage 1 than children who had been stuttering for more than 12 months. This suggests that older preschool children might require fewer clinic visits to complete Stage 1. This finding might be related to the smaller number of clinic visits obtained for

school-age children (Koushik et al., 2009). Finally, the number of clinic visits for the combined cohorts to complete Stage 1 was 11.

Summary

Further to the clinical trials evidence presented in Chapter 2, the Lidcombe Program is supported by treatment process evidence as described in this chapter. The program effects were found to be greater than those of natural recovery. Further evidence related to the psychological impact, social validity, long-term stability and speech and language behaviour of parents and children has demonstrated positive clinical outcomes. The program has been implemented for school-age children with positive outcomes. Further, the program has been translated and utilised in other countries including Germany and Saudi Arabia, where it was found to be effective with children from different cultural backgrounds.

The Lidcombe Program has been used increasingly by clinicians throughout the world. Thus, the Australian Stuttering Research Centre (ASRC) formed a consortium of highly trained members to deliver training to clinicians. The Lidcombe Program Training Consortium (LPTC) was established as an international group of members in eight countries who provide training in the delivery of the Lidcombe Program worldwide in both English- and non-English-speaking countries (Australian Stuttering Research Centre, 2009). Since 2001, more than 3,000 clinicians in North America have received a 2-day basic Lidcombe Program skills workshop. Although many clinicians in that region have been trained in the delivery of the Lidcombe Program, no large cohort retrospective recovery studies have been performed. Considering the potential of large number of clinicians using this approach in North America, such large cohort studies could be combined with the Australian and British file audits (Jones et al., 2000;

Kingston et al., 2003) to establish world-wide clinical benchmarks for the Lidcombe Program.

The file audits of the Lidcombe Program in Australia and the UK examined variables that might affect treatment time during Stage 1 of the Lidcombe Program. However, these studies did not evaluate whether the interval between clinic visits affected the duration of treatment. This information is important for several reasons. First, studies in Western Australia have shown that the demands for speech and language services are increasing (O’Leary, 2010) and therefore knowledge of the amount of required clinician time is essential for decisions regarding allocated clinic visits. Second, it is important that early stuttering be treated until the process is complete, in order to avoid the negative consequences of treatment failure. Therefore, optimal spacing between clinic visits is important in the decision. Finally, funding bodies for speech and language services require evidence of number of clinic visits and optimal spacing between visits to allocate appropriate monies and time for clients.

The next chapter describes a North American file audit of the Lidcombe Program that is a replication and extension of the Jones et al. (2000) study. This study is the first empirical research presented in this thesis. The file audit was conducted to evaluate the effects of predictor variables affecting treatment outcomes in a region where many clinicians are trained in the Lidcombe Program. Moreover, a new predictor variable that has not yet been explored, the time between clinic visits, is included in the evaluation to determine the relationship of time between clinic visits and treatment time during Stage 1.

Chapter 4

Lidcombe Program Outcomes in the Real World²

Lidcombe Program evidence was provided in the previous two chapters, with the focus on clinical trials evidence in Chapter 2 and treatment process evidence in Chapter 3. In the review of the literature, it was noted that predictor variables evidence was available in two large-group independent studies, one in Australia and the other in the United Kingdom (Jones et al., 2000; Kingston et al., 2003). In these studies, case variables from clinical files were examined from clinical communities. The purpose of both these studies was to determine whether the duration of treatment with the Lidcombe Program could be predicted. Further, the data from these studies were pooled for meta-analysis (Kingston et al., 2003). The meta-analysis improved benchmarking data for clinicians who use the Lidcombe Program.

Clinical benchmarking contributes knowledge about the process of clinical care and outcomes (Higgins, 1997). In speech- language pathology there is little published on clinical benchmarking (Hunt & Slater, 1999) and this is so for stuttering (Yaruss, LaSalle, & Conture, 1998). Benchmarking data enables healthcare professionals to compare treatment delivery to a standard. This allows for management of health services and allocation of funds for treatment approaches. Evidence-based practice is important in the evaluation of benchmarking data, to provide systematic research evidence for decision making.

In this study, case variables were examined in North-America, another region in which the Lidcombe Program is used extensively. This study is a replication and extension of the Jones et al. (2000) file audit, but another valuable variable, the time

² The following study has been published in the International Journal of Speech-Language Pathology and extended for this thesis (Koushik, Hewat, Shenker, Jones & Onslow, 2011).

between clinic visits, was added. The addition of the variable was to assess clinical translation of the Lidcombe Program. Further, the data from the North American, Australian and British studies were combined and meta-analysis performed to establish worldwide clinical benchmarks for the Lidcombe Program.

Method

Study Design

The procedures used by Jones et al. (2000) were replicated and extended in a retrospective file audit including four clinical sites from the United States and one site from Canada. The sites were chosen as the clinicians were known to routinely use the Lidcombe Program in those clinics. Personnel from the clinics extracted and de-identified requisite information from the files of all children who had been treated with the Lidcombe Program. Fifteen clinicians with varying levels of experience had treated the children with the procedures as described in the manual (Packman et al., 2010). All treating clinicians had received a 2-day Lidcombe Program basic skills workshop by members from the Lidcombe Program Training Consortium (Australian Stuttering Research Centre, 2009).

Ethics

Five speech and language clinics, one in Canada and four in the United States, were contacted for participation in this study. Consent was not required from the children who had completed Stage 1 or from their parents/caregivers, because the study was a retrospective file audit. Therefore, in each location the director of the clinic was deemed the “participant”, sent a participant information sheet, and was requested to sign a consent form for inclusion in this study. The consent form outlined confidentiality of participants’ identity, their voluntary participation and their freedom to withdraw from the research with no consequences. The University of Newcastle Human Research

Ethics Committee (UoN HREC) provided ethics approval on 9 June, 2009 (Approval No: H-2009-0086).

Subsequently, a co-researcher was assigned at each clinical site, who was permitted to access all files of children who had completed the Lidcombe Program between 2002 and 2009. Each file was de-identified and placed in a locked cabinet for access by the co-researcher at that site. The co-researcher reviewed all files and recorded file data and information on an Excel spreadsheet. This de-identified data was sent to the author for collation and analyses.

Participants

File data were collected from 165 children who had attended the clinics during the years 2002-2009 and had begun treatment when younger than 6 years. The number of clinical files contributed from each clinic was 54, 50, 31, 20 and 10. Children were included in the analyses if they had completed Stage 1, in order to provide clinical benchmarks for duration of clinic visits for the first stage. The criteria for Stage 2 entry in the Packman et al. (2010) manual are “(1) %SS less than 1.0 within the clinic, and (2) Severity rating (SR) scores for the previous week of 1 or 2, with at least four of these being 1” (Packman et al., 2010, p. 8). These criteria need to be achieved for three consecutive clinic visits.

Among the 165 children, non-progression to Stage 2 occurred in 27 cases (13.5%). That figure is important to mention as health care systems, funding bodies, clinicians and parents rely on such information for decision-making purposes. For the purpose of this study, however, these files were withdrawn from further analysis. Reported reasons for non-progression were that parent schedules conflicted with available clinic times so they were unable to continue with weekly treatment (10 cases), the child lost funding and could not continue on a private pay basis (4 cases), concurrent

pressing medical treatment (2 cases), the family felt that progress with the Lidcombe Program was slower than expected (5 cases), and no reason given (6 cases). Of the 27 children who did not progress to Stage 2, according to file data, 20 had decreased their stuttering severity by more than 2.0%SS from pre-treatment until the time of drop-out, three showed no change and four had missing file data at the time of drop-out.

The remaining 138 children, 105 boys and 33 girls, completed the first stage and progressed to Stage 2 of the Lidcombe Program. The number of children from each clinic included in the analysis was 46, 41, 27, 20 and 4 respectively. Data from one clinic were removed because only four children reached Stage 2 and it was felt that their data would not make a meaningful contribution to the analyses. Thus the final analyses were based on 134 children. Statistical analyses used were SAS for Windows, version 9.2 (SAS Institute, Cary, NC).

Variables

Dependent Variable

The dependent variable was the number of clinic visits required for entry to Stage 2 of the Lidcombe Program. This variable was categorised to represent short and long treatment duration. Short treatments were defined as fewer than 12 clinic visits; long treatments were 12 visits or more. Categorising the dependent variable was decided upon because treatment time as a continuous variable did not meet the requisite assumptions for least squares regression.

Predictor Variables

The following four predictor variables used by Jones et al. (2000) and Kingston et al. (2003) were obtained from each clinical file: gender, age at the first treatment visit, onset-to-treatment interval, and stuttering severity in %SS at the first treatment visit. Categorisation of the variables was as performed by Jones et al. Categorisation

avoided the assumption that any relationship with the dependent variable would be linear. Age at the first treatment visit was categorised into younger than 4 years and 4 years and older. Stuttering severity at the first treatment visit was categorised as less severe, being below 5.0%SS, and more severe, being greater than or equal to 5.0%SS. Onset to treatment interval—the time between stuttering onset and the first treatment visit—was categorised as shorter than 12 months and longer than 12 months. The latter categorisation reduced reliance on parent recall of the exact time of onset.

Although the Lidcombe Program manual specifies that treatment be provided with weekly clinic visits during Stage 1 (Packman et al., 2010), there are many reasons beyond a clinician's control why this may not occur. Failures to attend clinic appointments occur for various reasons, some of which are illness, scheduling conflicts or vacations. Moreover, two reports suggest that clinicians deviate from the weekly visit requirement in order to manage caseloads (O'Brian, Iverach, Jones, Onslow, Packman, Menzies, & 2011; Rousseau, Packman, Onslow, Dredge, & Harrison, 2002). Therefore, mean number of days between clinic visits was calculated for the cohort and found to be 11. This variable was categorised into frequent visits (fewer than 11 days) and infrequent visits (11 days or more).

Results

Analyses used were SAS for Windows, version 9.2 (SAS Institute, Cary, NC). Goodness-of-fit statistics were used to assess the final logistic models. Descriptive statistics for the predictor variables are presented in Table 4.1. The median age at the first treatment visit was 4.1 years ($SD=0.8$ years), median onset-to-treatment interval was 13 months ($SD=10.2$ months), median days between clinic visits was 10 ($SD=5.8$ days), and median %SS at the first treatment visit was 5.0%SS ($SD=5.1\%$ SS). The

median %SS at the first treatment visit was calculated for 131 instead of 134 clinic files because this information was missing from three files.

Table 4.1: Descriptive statistics for the North American data (N=134) (reproduced with permission)

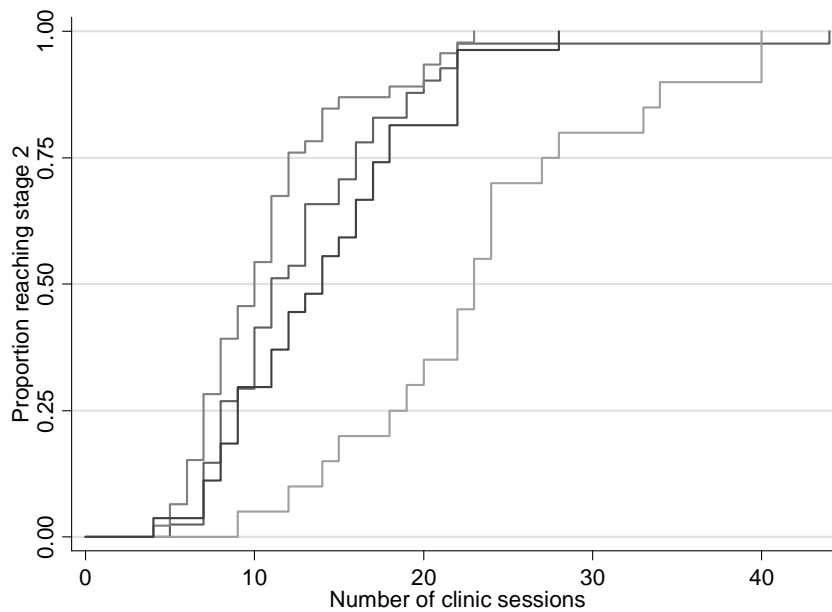
		Age at first treatment visit (months)	Onset to treatment interval (months)	%SS at first treatment visit	Days between clinic visits	Number of clinic visits to Stage 2	Number of clinic visits to Stage 2 (not including outlier)
N	Valid	124	122	131	132	134	114
	Missing	10	12	3	2	0	20
Mean		49.6	15.9	6.3	11	14.1	12.4
Median		49.5	13	5	10	12	11
Mode		51	8	3	10.5	7	7
Standard deviation		9.5	10.2	5.1	5.8	7.5	5.8
Range		31-71	1-53	0.3-32	2.3-49.9	4-44	4-44

Median Number of Clinic Visits by Clinic Site

As evidence of heterogeneity was found between the clinic sites (*logrank* $p=.01$), the analyses presented were stratified by clinic. A Kaplan-Meier survival analysis is a descriptive statistical procedure for the time-to-event variables (Kaplan & Meier, 1958). It is used in cases where time is the most prominent variable and involves the generation of survival plots. For the North American data, a survival analysis was used as a tool to measure the required time (i.e. measured in number of clinic visits) to reach Stage 2 (i.e. event). Figure 4.1 represents the cumulative proportion of children who attained Stage 2 by the number of clinic visits, stratified by clinic site. Median number of clinic visits is represented by 0.50 of the proportion of children reaching

Stage 2, or in other words, where 50% of all children reached near-zero stuttering. The median numbers of clinic visits required to attain Stage 2 were similar for all clinics except one. Medians for the clinics were 11, 10, and 14, and 23 visits for one clinic. Clearly, the outlying data for one clinic required further exploration. For the purposes of providing benchmarking data for number of clinic visits, the data from this clinic were included in the group analysis.

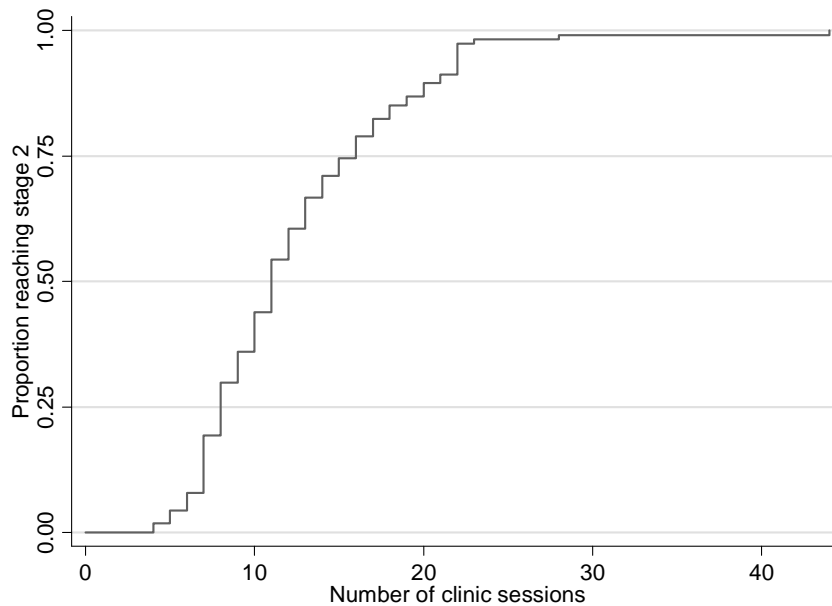
Figure 4.1: Kaplan-Meier plot of cumulative proportion of subjects who attained Stage 2 by clinic site (reproduced with permission)



Median Number of Clinic Visits for the Group

A Kaplan-Meier plot for the number of clinic visits is presented in Figure 4.2. For the 134 children, the median number of clinic visits was 12. The 90th percentile was 22 visits. If the files from the outlying clinic were not included in the analysis, the median and 90th percentile values decreased to 11 and 21 respectively. Four children were below 1.0%SS at the first clinic visit. To confirm that these children did not affect the median value for the whole cohort, the data were reanalysed without them. For the 130 files, the median number of clinic visits to Stage 2 remained at 12.

Figure 4.2: Kaplan-Meier plot of cumulative proportion of 134 subjects who attained Stage 2 by number of clinic visits (reproduced with permission)



Logistic Regression

To determine the relationship between the dependent variable and all five predictor variables, a univariable logistic regression analysis was performed. For predictor variables, one category was specified as the reference, and the non-reference value was measured for significance. The odds ratio is a measure of the strength of relationship between two variables. If the odds ratio for the non-reference value is 1.0 there is no difference between the groups. Table 4.2 shows the results of the univariable regression, stratified by clinic site.

The data showed no evidence of an association between number of clinic sessions and age, gender, or onset-to-treatment interval. There was strong evidence, however, that higher severity was associated with more clinic visits ($p=.004$). Children with stuttering severity of 5.0%SS or more had approximately four-fold increased odds of requiring 12 or more visits than the milder group. There was also some evidence that frequent clinic attendance was associated with more clinic visits to Stage 2 ($p=.04$).

Children who attended the clinic more often than every 11 days had more than twice the odds of requiring more than 12 clinic sessions than children who attended the clinic infrequently.

A multivariable logistic regression analysis showed similar results to the univariable analysis. The association between frequency of attendance and number of clinic sessions approaches statistical significance (odds ratio=0.47, $p=.07$). For the variable severity of stuttering, the association between severity and number of clinic sessions was almost unchanged.

Table 4.2: Results of univariable logistic regression for the North American data (reproduced with permission)

Variable		Odds Ratio	95% Confidence Interval	p-value
%SS at first clinic visit	<5%SS	1.0*		
	5%SS+	3.8	1.5 – 9.4	0.004
Onset-to-treatment interval	<12 mths	1.0*		
	12 mths+	1.1	0.50 – 2.6	0.8
Gender	Male	1.0*		
	Female	0.84	0.34 – 2.1	0.7
Age	<4 years	1.0*		
	4 years+	1.04	0.48 – 2.2	0.9
Attendance	More frequent	1.0*		
	Less frequent	0.44	0.20 – 0.96	0.04

* = reference category

Goodness-of-Fit

Goodness-of-fit statistics were used to assess the final logistic models. The c-statistic indicates how well a model distinguishes between children taking fewer and children taking more clinic sessions, where 0.5 indicates a model that is not predictive and 1.0 indicates a model that predicts perfectly. Pearson's chi-square test was used to

assess that the models did not provide a poor fit to the data. The final logistic model had a c-statistic of 0.79 and Pearson's chi-square = 10.4, df = 10, p = 0.4. These statistics indicated no evidence of lack of fit and a model that could reasonably distinguish between children taking more and children taking fewer clinic sessions.

Meta-Analysis

The data collection methods for the present study were identical to those of Jones et al. (2000) and Kingston et al. (2003). However, those studies did not collect data for number of days between clinic visits; therefore that variable was not included in the meta-analysis. The data sets for 444 children who attained Stage 2 of the Lidcombe Program for the three studies were combined. For the purpose of the meta-analysis, severity was re-categorised into three levels as speech pathologists often express stuttering severity as mild, moderate and severe. The categories of mild (0-4.9%SS), moderate (5.0-9.9%SS) and severe (10.0%SS+) were used to report findings. Results are presented in Table 4.3.

Table 4.3: Results of the univariable logistic regression (Australian, British and North American cohorts) (reproduced with permission)

Variable		Odds Ratio	95% Confidence Interval	p-value
%SS at first clinic visit	<5%SS	1.0*		
	5-9.9%SS	2.3	1.4-3.7	0.0008
	10%SS+	5.2	2.5-10.6	<0.0001
Onset-to-treatment interval	<12 mths	1.0*		
	12 mths+	0.76	0.50-1.1	0.18
Gender	Male	1.0*		
	Female	0.70	0.44-1.1	0.14
Age	<4 years	1.0*		
	4 years+	0.87	0.59-1.3	0.49

* = reference category

Based on meta-analysis of the 444 cases, there was no evidence of a correlation between age, gender, onset-to-treatment interval and treatment duration. However, there was strong evidence of correlation between stuttering severity and treatment duration. Based on the Kaplan-Meier analysis and log-rank test there was strong evidence that increasing severity was associated with increased number of clinic visits ($p < .0001$). For the group, the median number of clinic visits to Stage 2 was 11. Based on the re-categorisation into three levels, the median number of clinic visits to Stage 2 was 10 for mild, 12 for moderate, and 14 for severe pre-treatment severity of stuttering. More specifically, there was strong evidence that children with moderate pre-treatment severity had more than double the odds of a longer duration of treatment ($p = .0008$) than those with mild pre-treatment stuttering. Moreover, there was strong evidence that children with severe pre-treatment severity had more than five times the odds of longer duration of treatment ($p < .0001$) than those with mild pre-treatment severity of stuttering.

The final goodness-of-fit logistical model for the meta-analysis had a c-statistic of 0.67 and Pearson's chi-square = 13.4, $df = 12$, $p = 0.34$. These statistics indicate no evidence of lack of fit and a model that has some ability to distinguish between children taking fewer and children taking more sessions; however, a limitation is that frequency of attendance could not be included in the model.

Discussion

The present study replicated the methodology of Jones et al. (2000), with the addition of a new predictor variable, and combined the data sets with the Australian and British studies for meta-analysis to provide benchmarking data. Of the five participating North American clinics, one was removed from the analysis because of the small number of contributed files. The median visits to complete Stage 1 by clinic site were

similar for three clinics at 11, 10 and 14. However, the fourth clinic reported a median of 23 visits. The difference for this clinic could not be explained by higher severity of stuttering. Possible explanations could be differences in the service delivery of the Lidcombe Program, differences in clinician experience, or data errors. To provide benchmarking data for clinic visits to Stage 2, the files from this clinic were included in the final analysis.

Pre-treatment stuttering severity was found to be a significant predictor of treatment time for the North American cohort. Stuttering severity of 5.0%SS or higher required more sessions to complete Stage 1 than did lower pre-treatment severity. However, the three predictor variables onset-to-treatment interval, gender, and age at first treatment visit were not found to be significant predictors of treatment time. The variable frequency of clinic visits produced an unexpected finding. On average, children who attended the clinic frequently (averaging fewer than 11 days between visits) required more clinic visits to complete Stage 1 than did infrequent attendees (averaging 11 days or more between visits). A statistical trend in the multivariable regression showed some evidence of an association ($p=.07$), although marginally so. This is a clinically important trend in the data that requires further investigation because of its potential clinical significance.

The number of clinic visits to Stage 2 was similar to the numbers reported by the Jones et al. (2000) and Kingston et al. (2003) studies. In these studies, the reported median value of 11 visits was similar to the North American cohort which reported 12 clinic visits. Further, the 90th percentile value for the North American cohort was 22 clinic visits. In comparison, 90% and 95% of the Australian and British cohorts completed Stage 1 in 22 and 21 clinic visits, respectively. All three studies independently agreed on the median and 90th and 95th percentile values. For treatment

time to Stage 2, the Australian and British studies did not find significant results for gender and age at first treatment visit. These results were similar to those obtained from the North American cohort.

The meta-analysis of the three studies increased the statistical power, thus providing important benchmarking information. A highly significant predictor for treatment time was pre-treatment severity, which was re-classified into three categories. The median number of clinic visits for different severity was 10 for mild, 12 for moderate and 14 for severe pre-treatment stuttering severity. The meta-analysis by Kingston et al. (2003) showed a significant correlation between onset-to-treatment interval and treatment time. However, when the North American data were included, this correlation became non-significant. A test for interaction was used to determine whether there was a differential effect in the Kingston et al. data compared to the North American data. However, this was not found to be the case (Wald Chi-Square 1.25, $df = 1$, $p = 0.26$).

An important finding was the agreement of median values obtained from the North American cohort independent of the Australian and British cohorts. Further, the meta-analysis provided important benchmarking data for clinical translation. It is important to note that the Australian, British and North American studies were performed in primarily English-speaking nations. Investigations of the Lidcombe Program with non-English speaking countries are required to determine whether these benchmarks are achievable in countries with different languages and cultures.

The Lidcombe Program manual specifies that treatment be provided on a weekly basis during Stage 1 clinic visits (Packman et al., 2010). However, closer examination of the North American data revealed that weekly clinic visits were not achieved and in fact clients attended on average every 11 days. Of the 27 participants who did not

complete the first stage, 10 (37%) non-completions were attributed to parents' inability to continue with weekly clinic visits due to scheduling conflicts. Failures to attend clinic appointments occurred for various other reasons, including illness or holidays. The finding of attendance frequency shows that the Lidcombe Program is not being followed as per the manual and a translation problem is encountered. Further, two reports have suggested that clinicians deviate from the weekly visit requirement in order to manage caseloads (O'Brian, Iverach, Jones, Onslow, Packman, et al., 2009; Rousseau, et al., 2002). Therefore, to suit health care practitioners and parents who cannot manage weekly treatment, offering alternative treatment schedules might be necessary. To date it is unknown how altering weekly clinic visits of the Lidcombe Program might affect treatment efficacy and efficiency.

Although the findings from this study suggested that weekly treatment was not being achieved in clinical communities, the findings from the North American file audit suggested that clinic visits more than 7 days apart might be more efficient than the standard. It was found that a longer time between clinic visits (average 11 days or more) resulted in fewer clinic visits than a shorter time between clinic visits (average less than 11 days). Although the findings suggested that the efficiency of the Lidcombe Program could be improved, it is not known whether the efficiency gained was at the expense of the efficacy of treatment outcomes. Thus there is a need for a well designed clinical trial examining the effects of different clinic visit schedules on treatment efficiency and efficacy. The following chapter is the second empirical study of this thesis, designed to add to the evidence base by examining the effects of different treatment schedules on both the efficiency and efficacy of the Lidcombe Program.

Chapter 5

Does Changing the Frequency of Lidcombe Program Clinic Visits Affect Outcomes?

The need for speech and language services is increasing worldwide and clinicians are managing larger caseloads of children. In parts of Australia, waiting times for treatment have increased up to 16 months (O’Leary, 2010). Studies in Canada and the United Kingdom highlight the concerns of both professionals and parents regarding waiting times for child developmental and rehabilitation services (Clow, Mustafa, Szollar, Wood, Reid, et al., 2002; Keating, Syrmis, Hamilton & McMahon, 1998). The area of concern is validated by the increasing body of evidence supporting early intervention and its benefits for long-term treatment outcomes (Bloodstein & Bernstein Ratner, 2008; Reilly et al., 2009). Different methods for managing waiting lists have been trialled in the hope of resolving this issue. In a United Kingdom speech and language clinic, the service delivery model was re-organised from individual to group treatment (Miller, Armstrong, Masse, Klassen, Shen, et al., 2008). Rather than one-on-one therapy, the emphasis was placed on parent training in groups. The change in service delivery resulted in decreased waiting times for both assessment and treatment.

In stuttering, weekly treatment with the Lidcombe Program is recommended in the Lidcombe Program manual (Packman et al., 2010). Rousseau and colleagues (2002) designed a questionnaire to determine whether clinicians were adhering to the Lidcombe Program manual in Australian general clinics. A questionnaire was sent to 400 randomly selected clinicians regarding their practice of the Lidcombe Program. Of the 277 respondents, 154 clinicians (87%) reported using the Lidcombe Program in their practice. However, only half indicated that they delivered the program as per the manual. Of all respondents, only 50% adhered to weekly therapy sessions; it could be

assumed, therefore, that the others delivered treatment on a different schedule. The respondents reported that the main treatment delivery barriers relating to the workplace were large caseloads, inability to offer the intensity and length the treatment required or to allocate the necessary time. In a similar study, Shenker, Hayhow, and Lawlor (2005) used a web-based questionnaire to determine clinicians' attitudes regarding Lidcombe Program treatment. The study surveyed 213 trained clinicians in Canada, of whom only 50% indicated that they followed the manualised procedures in its researched format. Difficulties for implementation of the program by clinicians were related to the length of time required for children to complete Stage 1. Thus both studies provided evidence that administration of the Lidcombe Program in general clinics was not the same as it would be in efficacy trials.

The above studies were conducted by questionnaire and therefore the findings were based on clinician reports. To determine the extent to which clinicians followed the manual in general clinics, O'Brian and colleagues (2011) followed the normal practices of clinicians in a prospective study. The participants were 31 speech pathologists treating 57 children with the Lidcombe Program in public and private general clinics in Australia. The therapists were asked to deliver the Lidcombe Program as usual and not to change the way treatment was conducted. At the completion of the Stage 1 for each child, the speech pathologists provided the researcher with details regarding each child's clinical progress by using a checklist. Results showed that 49% of clinicians were likely to schedule 30 minute therapy sessions rather than the 45-60 minutes suggested in the manual. The average time between clinic visits was found to be 15 days, just over a fortnight, rather than the 7 days suggested in the manual (Packman et al., 2010). Some workplaces did not allocate weekly visits due to large caseloads; therefore, scheduling of appointments was adjusted by the clinical institution.

Further, sessions were missed by families due to cost of treatment, busy lifestyle, and work commitments.

In the previous chapter, a file audit study was performed in North American clinics. The file audit examined predictor variables of children who had completed Stage 1 of the Lidcombe Program. The methodology was a replication of that used by Jones et al. (2000), with the addition of a new predictor variable, frequency of clinic visits. The new variable was added to determine the average time between clinic visits. The predictor variables were measured against the dependent variable, number of clinic visits to complete Stage 1. Evidence of non-adherence to weekly treatment was found. The average time between clinic visits was 11 rather than 7 days. The finding shows that clinical translation of the Lidcombe Program manual was not achieved. This finding was similar to that of O'Brian and colleagues (2011), thus providing evidence that weekly treatment schedules were not met. Moreover, there was some evidence that frequent clinic attendance was associated with increased number of Stage 1 clinic visits ($p = .04$). Children who attended the clinic more often than every 11 days had more than twice the odds of requiring more than 12 clinic visits compared to those who attended the clinic less frequently. This was a surprising finding that was investigated in the present prospective empirical study. As of yet, no research has confirmed whether scheduling treatment visits at intervals greater or less than a week is as efficient or efficacious as the standard Lidcombe Program.

The present study was a Phase II clinical trial of the Lidcombe Program, varying the service delivery model of treatment. The purpose was to evaluate the effects of different treatment schedules (i.e. number of days between clinic visits) on the treatment efficiency and efficacy of the Lidcombe Program.

Method

Study Design

This study was a Phase II clinical trial of three service delivery models of the Lidcombe Program. The purpose of a Phase II design is to measure the response of different experimental groups to the treatment schedules, rather than establishing whether the treatment is effective (Hackshaw, 2009). According to Onslow and colleagues' (2008) definition of a clinical trial, the present study is classified as Phase II rather than Phase I, as the trial was not a preliminary investigation of a new treatment and subject numbers were greater than 10. Further, the trial is not classified as a Phase III clinical trial as the subject numbers were not in the range of several hundreds to thousands. The study meets the clinical trial criteria as it comprised the following three features. First, the study included an entire treatment, as outlined in the Lidcombe Program manual (Packman et al., 2010). Second, outcome measures were available in percentage of syllables stuttered (%SS) and severity ratings (SR). Finally, the outcome measures were evaluated by a blinded observer, a speech pathologist independent of the study.

Ethics

Ethics clearance was obtained from The University of Newcastle Human Research Ethics Committee (HREC) for the conduct of this study. Ethics approval was granted on 13th December 2006 (Approval No: H-346-1206) to commence the clinical trial at the Montreal Fluency Centre in Canada and recruitment commenced January 2007. A variation to the approval was submitted to the HREC to add the University of Newcastle Stuttering Clinic as a second research site. Approval was granted on the 13th February 2008 and recruitment commenced in March 2008. All interested participants were provided with a detailed information sheet outlining the requirements of the

research. Written consent to participate was obtained from the families before they entered the research study.

Research sites

Participants were recruited from the waiting list at specialised stuttering clinics in Australia and Canada. The two sites were the stuttering clinic at the University of Newcastle, Australia and The Montreal Fluency Centre in Montreal, Canada. The sites were chosen because both clinics employed clinicians who specialised in treatment of early stuttering. Two clinicians, one at each site, provided all treatment for this research. The treating clinicians had both received previous training by the Lidcombe Program Training Consortium (LPTC) and were experienced in the delivery of the Lidcombe Program. One clinician was the author of this thesis and the second an employee of the Montreal Fluency Centre. The author was responsible for all data collection and management at both research sites.

Randomisation

Children were randomly allocated into one of three service delivery groups. The control group received standard weekly visits and the two experimental groups received treatment twice weekly (intensive treatment schedule) or fortnightly (less intensive treatment schedule). Randomisation ensured that the groups were as similar as possible except for the variable of interest. Furthermore, stratified randomisation ensured that the groups were similar across certain characteristics, so as not to influence the response to the intervention.

The first variable accounted for in stratification was the severity of the speech and/or language disorder. Rousseau and colleagues (2007) found that receptive language scores and mean length of utterance (MLU) correlated significantly with treatment time during Stage 1. Therefore, this variable was accounted for in

stratification so that each group had similar severities of speech and/or language disorder(s). The second variable was the severity of stuttering measured in %SS. It is known that higher pre-treatment severity (5.0%SS or more) inflates treatment time with the Lidcombe Program to more than a median 11 clinic visits (Jones et al., 2000; Kingston et al., 2003; Koushik, Hewat, Shenker, Jones, and Onslow, 2011). Stratification ensured that the groups had similar median stuttering severities. The final stratification variable was treatment site. This was to ensure that the groups from both sites, Newcastle and Montreal, had children with similar characteristics.

Inclusion and Exclusion Criteria

A stuttering assessment was conducted by the treating clinician after families had consented to participate in the research. During the initial assessment a diagnosis of stuttering was confirmed by consensus between the parent and clinician. Inclusion criteria required that the child had been stuttering for longer than 6 months, had had no previous intervention with the Lidcombe Program, and both the parent and child had functional levels of English. Furthermore, the child's stuttering level was greater than 2%SS, confirmed by at least one recording of the child's speech obtained in a speaking situation beyond the clinic. Children were excluded if they were stuttering less than 2%SS, if parents reported a diagnosis of attention deficit hyperactivity disorder and/or intellectual disability, or if the child was diagnosed with severe speech and/or language disorder. Speech disorders were diagnosed by the Diagnostic Evaluation of Articulation and Phonology (Dodd, Hua, Crosbie, Holm, & Ozanne, 2006). Language disorders were diagnosed by the Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Semel, Wiig, & Secord, 2004)

To obtain the beyond clinic (BC) speech samples, each family was provided with an audio tape recorder and a blank tape to record the child's speech in natural

speaking environments after the initial assessment. Parents were asked to audio-tape two 10-minute conversations. One conversation was recorded in the home environment, with the child talking with the mother or father. The other conversation was recorded outside of the home, with the child talking with a teacher or grandparent. The parent returned the audiotape to the treating clinician who rated the two conversation samples for %SS using a two-button rating machine.

Once the child met all requirements for inclusion in the study, a unique identification number was assigned to each participant by the treating clinician at the University of Newcastle stuttering clinic or at the Montreal Fluency Centre. The child's identification number, treatment site, speech and language diagnosis categorised as mild or moderate, and stuttering severity in %SS were sent to an independent researcher associated with the Australian Stuttering Research Centre (ASRC) at the University of Sydney. This researcher performed the randomisation procedures for all children included in this study and notified the clinician in Montreal or Newcastle of the treatment group for each child, that is, weekly, twice weekly or fortnightly.

Treatment

All children received the Lidcombe Program of early stuttering intervention as outlined in the manual (Packman et al., 2010) and described in Chapter 3. The exception to the manual was the scheduling of clinic visits for the two experimental groups. Children randomised into the fortnightly or twice weekly groups received treatment on those schedules. To ensure consistency between clinicians and to avoid other potential influences on treatment outcomes, the research protocol highlighted three additional treatment procedures. These included 1) minimal contact between clinic visits, 2) clinic visits to remain between 45 to 60 minutes, and 3) clarification on criteria for entry to Stage 2. For each treatment group, entry into Stage 2 was determined as per the

Lidcombe Program manual (Packman et al., 2010). The criteria for entry to Stage 2 are three consecutive clinic visits where the child's speech is rated less than 1.0%SS and SR scores for the previous week of mostly 1's (where 1 = no stuttering, 2 = extremely mild stuttering and 10 = extremely severe stuttering). However, due to the different clinic visit schedules, the number of days preceding entry to Stage 2 differed for all three groups. In the fortnightly group, children entered Stage 2 after no fewer than 35 days, in the standard group after no fewer than 21 days and in the twice weekly group after no fewer than 14 days. In Stage 2, all children were seen for regular maintenance visits at decreasing frequency, as recommended in the manual.

Participants

The participants were 31 stuttering preschool children, between the ages of 3 years 0 months and 5 years 11 months on the date of assessment. Of the 31 children, 26 were boys and five were girls. Eight children were allocated to the standard weekly group, 11 to the twice weekly and 12 to the fortnightly group. The median age was 4 years 2 months (range 3 years 0 months – 5 years 6 months). The median BC stuttering severity for the 31 children was 5.9%SS. In each treatment schedule, the median %SS was 7.0 for weekly, 5.5 for twice weekly and 5.5 for fortnightly, respectively. No child had received previous therapy for stuttering. Fifteen children had a positive family history of stuttering and 12 presented with other speech and/or language disorders besides stuttering. Of those 12, eight had a mild language delay, one had a mild-moderate language delay, one had a moderate language delay, one had a moderate phonological delay and one had both mild-moderate language and moderate phonological delay. A summary of the participants by group is presented in Table 5.1. Details of all participants are presented in Appendix Table A.1.

Table 5.1: Summary of recruited participants by group

Detail	Weekly	Twice Weekly	Fortnightly
No. of participants	8	11	12
Median age	4 years 0 months	4 years 4 months	4 years 0 months
Median stuttering severity (%SS)	7.0%SS	5.5%SS	5.5%SS
No. of participants with a family history of stuttering	3	5	7
No. of participants with other speech and language disorders	3	4	5

Drop-outs and Withdrawal

For ethical reasons, the research protocol stated that children who did not meet Stage 1 criteria by 6 months in either of the two experimental groups (fortnightly or twice weekly) must be withdrawn by the researchers and provided with standard weekly sessions. Of the 31 children who were randomised into treatment groups, altogether 10 were withdrawn or independently dropped out of the research, two before and eight after commencing treatment. The two children who dropped out before commencing treatment were from the twice weekly group. Of the eight children who were withdrawn or dropped out of the research during treatment, three were from twice weekly, one from weekly and four from the fortnightly groups. A summary of these 10 children are provided in Table 5.2.

In the weekly group, one child (participant 8) dropped out after 29 clinic visits due to the parents' concern regarding the child's sensitivity. The parents felt that the child was reacting negatively to any feedback, both at home and in the clinic. The

extreme sensitivity was not only associated with verbal contingencies from the program but was apparent in unpredictable random conversation.

In the twice weekly group, two children dropped out (participants 15 and 17) before commencing treatment due to the parents being unable to schedule two clinic visits each week. The parents of one child (participant 16) dropped out after receiving 20 twice weekly visits as they could not continue attending the clinic twice a week. This family was offered treatment on a weekly schedule but decided not to continue as the available treatment times of the clinician were not suitable. Two children were withdrawn (participants 18 and 19) from the twice weekly group after 56 and 45 clinic visits, due to research protocol. Both these families were offered weekly treatment, but decided not to continue.

In the fortnightly group, one child dropped out after 4 clinic visits (participant 31) due to family relocation. Another child dropped out after 10 clinic visits (participant 30) because the parent and speech pathologist felt that this child needed more clinical support as they were not seeing the expected progress. This child received weekly treatment until the completion of Stage 1 in a total of 66 clinic visits. Two additional children (participants 28 and 29) were withdrawn from the fortnightly group after 16 and 17 clinic visits due to the study protocol and received weekly sessions. Both children eventually completed Stage 1 in a total of 20 and 30 clinic visits respectively.

Table 5.2: Details of participant drop-out or withdrawal

<i>Participant Number</i>	<i>Group</i>	<i>Stage Reached</i>	<i>Clinic Visits</i>	<i>Clinic Visits to Stage 2</i>	<i>Total Weeks</i>	<i>Pre-treatment (%SS)</i>	<i>%SS at D/O* or W/D</i>	<i>Reason for Drop-out or Withdrawal</i>
8	Weekly	D/O	29	-	35	8.5	0.7	Sensitive child
15	Twice weekly	D/O	0	-	-	3.2	-	Scheduling
16	Twice weekly	D/O	20	-	14	4.3	2.8	Scheduling
17	Twice Weekly	D/O	0	-	-	5.4	-	Scheduling
18	Twice weekly	D/O	56	-	40	33.3	1.7	Required by protocol/ then dropped out
19	Twice weekly	D/O	45	-	28	6.0	3.2	Required by protocol/ then dropped out
28	Fortnightly	STAGE 2	16	20	49	6.5	1.2	Required by protocol/ then completed
29	Fortnightly	STAGE 2	17	30	60	7.7	0.6	Required by protocol/ then completed
30	Fortnightly	STAGE 2	10	66	100	7.5	5.3	More support required
31	Fortnightly	D/O	4	-	8	3.0	0.9	Moved

*D/O=drop-out, W/D=withdrawal

Participants Who Completed Stage 1

Overall, 21 children completed the first stage in their allocated treatment schedule, seven in the weekly, six in the twice weekly and eight in the fortnightly groups. Of these children, 18 were boys and three were girls. Eleven children had a positive family history of stuttering. The number of children who completed treatment in Newcastle and Montreal were 14 and seven respectively. Ten children presented with concomitant speech and/or language disorders including three in the weekly, three in the twice weekly and four in the fortnightly groups. The median age at assessment, median

pre-treatment stuttering severity and other details of children who completed Stage 1 in their allocated treatment schedule are presented in Table 5.3.

Table 5.3: Details of children who completed Stage 1 in treatment schedule

Detail	Weekly	Twice Weekly	Fortnightly
No. of children completing Stage 1	7	6	8
Gender	7 boys	5 boys 1 girl	6 boys 2 girls
No. of children with a positive family history of stuttering	3	3	5
No. of children at each treatment site	4 Newcastle 3 Montreal	4 Newcastle 2 Montreal	6 Newcastle 2 Montreal
*No. of children with concomitant speech and language disorders	2 mild language 1 mod. language	2 mild language 1 mod. phono & mild-mod. lang	2 mild language 1 mild-mod. lang 1 mod phono
Median age at assessment	4;3 years	4;7 years	4;8 years
Median pre-treatment stuttering severity (%SS)	7.0	5.5	5.5

*mod=moderate, phono=phonology, lang=language

Outcomes

Various data were collected to determine the outcome of treatment for each participant and to compare outcomes across the three treatment groups. Individual data were collected for each child and recorded on a Microsoft Excel spreadsheet. The data sheet documented the session date, missed sessions in the allocated schedule, number of clinic visits, session %SS and average SR for the previous week. Time spent consulting with the parent between clinic visits was also recorded. These data were used to record outcome variables and to track individual progress with the Lidcombe Program.

The outcomes of this clinical trial were determined in two ways, by evaluating the effects of the different treatment schedules on (a) the efficiency and (b) the efficacy of the Lidcombe Program. The efficiency of the treatment schedules was the measurement of the rate of completion of Stage 1 for all three groups. This was determined by two outcome measures. The primary outcome measure was the number of clinic visits and secondary outcome measure was the number of weeks to complete Stage 1. Efficacy of the treatment schedules was determined by evaluating the severity of stuttering of the three groups at different assessment occasions and comparing the results to the previous research (Jones et al., 2005).

Efficiency of the Treatment Schedules

The number of clinic visits to complete Stage 1 was the primary outcome for this Phase II clinical trial. This measure was obtained by counting the number of clinic visits that the child attended from the first visit to completion of program criteria. The number of weeks to complete Stage 1 was the secondary outcome measure. This was obtained by subtracting the date of the final treatment session from the date of the first treatment session, which recorded the number of days. To calculate the number of weeks for each child to complete Stage 1, the total number of days was divided by seven. The data for number of weeks were recorded on an individual Excel spreadsheet for each child.

Efficacy of the Treatment Schedules

The efficacy of the treatment schedules was determined by measuring stuttering severity at different occasions and comparing results to previous research. The %SS and the SR were collected for each child on four different assessment occasions: pre-randomisation, end of Stage 1, 9 months post-randomisation and 18 months post-randomisation. Nine months post-randomisation was chosen because findings could be compared to previous efficacy research of a Phase III RCT of the Lidcombe Program

(Jones et al., 2005). The comparison of groups at 9 and 18 months post-randomisation allowed for measurements of long-term outcomes and stability of outcomes when different dosages of clinic visits were delivered. On each assessment occasion, samples of the child speaking within and beyond the clinic were obtained. At pre-treatment, 9 months post-randomisation and 18 months post-randomisation, one within-clinic (WC) and two beyond-clinic (BC) samples were obtained. At the end of Stage 1, a WC sample was obtained. All WC speech samples were collected during a conversation between the child and clinician and were video-recorded on a Panasonic hand-held recorder (NV-GS300). All BC samples were obtained in the child's everyday speaking environments, as previously described. Altogether, 10 conversational samples of 10-minutes' duration had been collected for each child. A summary of the speech sampling frequency over time is presented in Table 5.4.

Table 5.4: Number of within- and beyond-clinic recordings required on each assessment occasion

	Occasion 1	Occasion 2	Occasion 3	Occasion 4
	Pre-treatment	Entry to Stage 2	9 months post-randomisation	18 months post-randomisation
Within-clinic video	1	1	1	1
Beyond-clinic audio	2	0	2	2

Stuttering Severity

The severity of stuttering was determined by recording two measures, %SS and SR, as described in Chapter 3. Severity ratings are typically used by parents during the administration of the program but have also been used to provide outcome data for stuttering interventions. Recent research suggests that SR may be equally reliable to %SS measures, except in the case of a high rate of repetitions or low rate of significant fixed postures without audible airflow (O'Brian, Packman, Onslow, & O'Brian, 2004).

In those cases, the authors suggest that a combination of the two measures would provide a valid assessment of a person's stuttering.

Of the 240 speech samples of children who completed Stage 1 in this study (21 children in their allocated treatment schedule + three who were withdrawn and then completed, x 10 speech samples), 195 (81%) were obtained by the researchers. The missing WC and BC ratings resulted from unavailability of participants to attend the clinic for WC video-recordings (N=11) and/or BC audio-tapes not returned to the researchers (N=34). The number of collected WC and BC speech samples on each assessment occasion is shown in Table 5.5.

Table 5.5: The total number of collected WC and BC speech samples on each assessment occasion

	Occasion 1	Occasion 2	Occasion 3	Occasion 4
	Pre-treatment	Entry to Stage 2	9 months post-randomisation	18 months post-randomisation
Within-clinic video	24	23	20	18
Beyond-clinic audio	48	0	40	22

A speech pathologist who specialised in stuttering but was independent of the research was recruited as a blinded observer to rate each sample for %SS and SR. The speech pathologist listened to the audio recordings on the listening tapes with a Sony Cassette Recorder (TCM-939) and Sony earphones, and watched the video recordings on a Macintosh laptop computer. A two-button press counting device called the True Talk was used to calculate %SS for both audio and video samples. After recording the %SS score, the speech pathologist also recorded a SR for all audio and video samples. Both measures were recorded on a data sheet and sent to the author for collation and analysis. The BC %SS and SRs reported are an average of the two collected samples at

each assessment occasion. The BC individual outcomes for the 24 children who completed Stage 1 of the Lidcombe Program are presented in Appendix Table A.2.

Inter-Judge and Intra-Judge Reliability

To establish inter-judge and intra-judge reliability for %SS and SR measures, 10% of the entire set of 240 speech samples was re-rated. To establish intra-judge reliability, the same blinded observer re-measured 24 randomly selected speech samples, 2 months after having rated the original samples. To measure inter-judge reliability, a second speech pathologist who specialised in stuttering treatment and was independent of the study rated the same 24 random samples. Intra-class correlation (ICC) was used to measure the consistency of measurements by different observers measuring the same quantity. For %SS and SR respectively, ICCs were 0.98 and 0.92 for intra-judge and 0.70 and 0.84 for inter-judge scores. Thus, the %SS and SR scores were considered reliable.

Parent Questionnaire

After completion of the research, all participating families were given a post-treatment questionnaire. From an EBP perspective, the information gathered from the questionnaire provided insight into patient perspective regarding treatment schedules. The questionnaire was given after the child had completed Stage 1 in the allocated schedule or if the child was withdrawn or dropped out of the research. The families were encouraged to return the questionnaire by a self-addressed stamped envelope to the author for collation and analysis. No identifying information was present on the questionnaire and all responses were kept confidential. The purpose was to determine the parents' likes and dislikes about the program and the treatment schedule to which they had been randomly allocated. The first three questions asked parents to provide a rating on a scale of 1-10. All other questions were open-ended, allowing the parents to

answer the questions freely. The questions of interest are discussed in detail in the results section. The questions were:

1. On a scale of 1-10, where 1= no stuttering and 10= extremely severe stuttering, what is the typical average severity rating of your child's speech during the last week?
2. On a scale of 1-10, where 1= no stuttering and 10= extremely severe stuttering, what is the most severe severity rating of your child's speech during the last week?
3. On a scale of 1-10, where 1=extremely satisfied and 10=totally dissatisfied, how satisfied are you with your child's present level of fluency?
4. Did you like the Lidcombe Program?
5. What did you like about the Lidcombe Program?
6. Did you have enough clinical support?
7. Was there enough progress between clinic visits?
8. If not, why not?
9. Did you have enough confidence to carry out treatment activities at home?
10. Did you think that the frequency of clinic visits was appropriate?
11. Why or why not?
12. If you were to start over and choose an ideal frequency of clinic visits, what would you choose?
13. Was there anything challenging?
14. Other comments

Results

Efficiency of Treatment Schedules

Primary Outcome

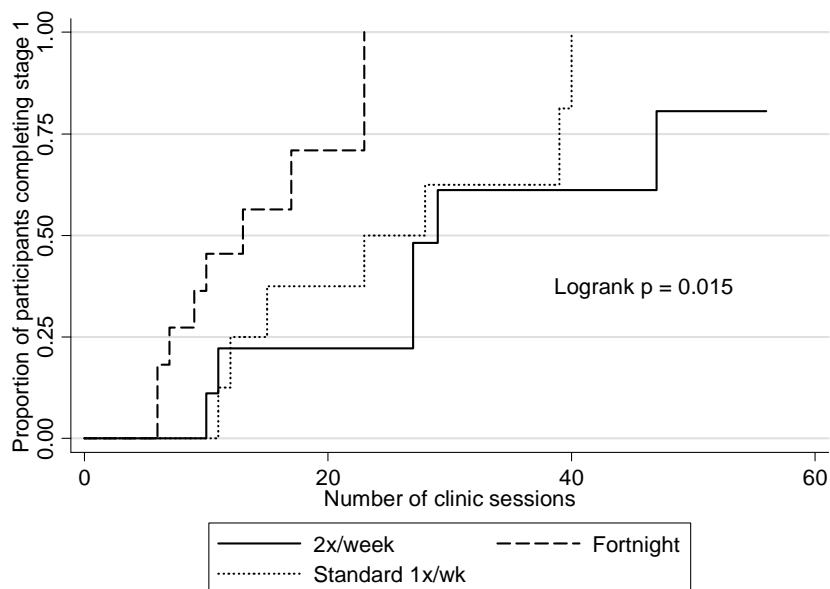
The primary outcome measure was the number of clinic visits to complete Stage

1. A stratified Cox regression procedure was used to determine association of the

treatment schedules with clinic visits to complete Stage 1. Analyses used SPSS Statistics 17.0 (SPSS 17.0, 2008). Participants who dropped out or were withdrawn from their allocated schedule (N=10) were “censored”. That means that data for these children were included until the time of departure from the schedule; therefore clinic visits were included until that point.

A Kaplan-Meier plot was used to show patterns of recovery within the cohort. A description of the Kaplan-Meier plot was provided in Chapter 4. As evidence of heterogeneity was found between the treatment groups (*logrank* $p=.002$), the presented analyses were stratified by treatment schedule. Figure 5.1 presents the cumulative proportion of children who attained Stage 2 by the number of clinic visits stratified by weekly, twice weekly and fortnightly sessions.

Figure 5.1: The cumulative proportion of children who attained Stage 2 by the number of clinic visits, stratified by weekly, twice weekly and fortnightly sessions.



A stratified Cox regression procedure determined that the median number of clinic visits to complete Stage 1 by treatment schedule was 23 for weekly, 27 for twice weekly and 10 for fortnightly sessions. The findings showed a significant difference between treatment groups for the primary outcome, clinic visits ($p = .01$). No significant

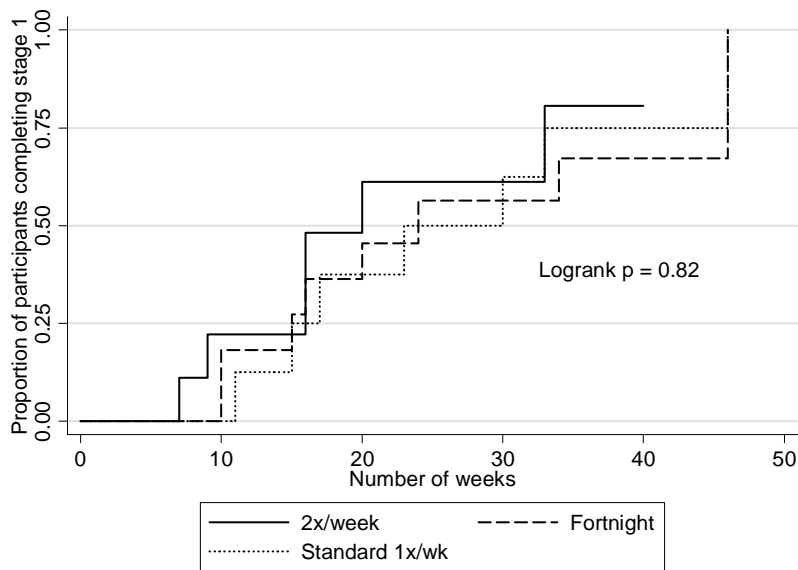
difference was found between twice weekly and weekly treatment schedules ($p = .28$), but a significant difference was found between fortnightly and weekly treatment schedules ($p = .02$). The effect size showed that children in the fortnightly group were 3.8 times more likely to have reduced clinic visits than those in the weekly group.

Secondary Outcome

The secondary outcome measure was the number of weeks to complete Stage 1. As with the primary outcome measure, a stratified Cox regression procedure was used to determine association of the treatment schedules with number of weeks to complete Stage 1. Participants who dropped out or were withdrawn from their allocated schedule ($N=10$) were censored. Thus data for those children were included until the time of departure from the schedule; therefore the number of weeks were included until that point.

A Kaplan-Meier recovery plot for number of weeks to complete Stage 1 found no significant difference between the groups (*logrank* $p=.748$). Figure 5.2 presents the cumulative proportion of children who completed Stage 1 by number of weeks, stratified by weekly, twice weekly and fortnightly sessions. The median numbers of weeks by treatment schedule were 18 for fortnightly, 23 for weekly and 16 for twice weekly sessions.

Figure 5.2: The cumulative proportion of children who completed Stage 1 by the number of clinic visits, stratified by weekly, twice weekly and fortnightly sessions



Efficacy of Treatment Schedules

Stuttering Severity

Findings from BC measures could be compared to previous efficacy research which employed a similar methodology (Jones et al., 2005). When the data from the different treatment schedules of the present study were combined, the median %SS at each assessment occasion were 8.2 pre-treatment, 1.9 at 9 months post-randomisation and 1.2 at 18 months post-randomisation. The median SR at each assessment occasion was 5.5 pre-treatment, 2.0 at 9 months post-randomisation and 2.0 at 18 months post-randomisation. In a Phase III RCT of the Lidcombe Program, the mean %SS for 29 children at 9 months post-randomisation was 1.5 (Jones et al., 2005), which is similar to the overall results obtained.

By treatment schedule, the median BC %SS in the twice weekly group was 10.3 pre-treatment, 2.4 at 9 months post-randomisation and 1.0 at 18 months post-randomisation. In the weekly group, the median %SS was 9.0 pre-treatment, 2.0 at 9 months post-randomisation and 0.7 at 18 months post-randomisation. Finally, in the

fortnightly group, the median %SS was 7.9 pre-treatment, 1.0 at 9 months post-randomisation and 1.4 at 18 months post-randomisation. In comparison to the results of Jones et al. (2005), the 9-months post-randomisation data were similar for the fortnightly group; therefore treatment efficacy did not appear to be affected by this treatment schedule.

The median BC SR in the twice weekly group was 6.8 pre-treatment, 2.5 at 9 months post-randomisation and 2.0 at 18 months post-randomisation. In the weekly group, the median SR was 5.5 pre-treatment, 2.3 at 9 months post-randomisation and 1.5 at 18 months post-randomisation. Finally, in the fortnightly group, the median SR was 5.0 pre-treatment, 2.0 at 9 months post-randomisation and 1.6 at 18 months post-randomisation. Table 5.6 presents the median WC and BC stuttering severity data by treatment schedule at pre-treatment, entry to Stage 2, 9 months post-randomisation and 18 months post-randomisation.

Table 5.6: Median WC and BC %SS (SR) by treatment schedule at pre-treatment, entry to Stage 2, 9 months post-randomisation and 18 months post-randomisation.

Treatment Schedule	Within or beyond clinic	Pre-treatment	Entry to Stage 2	9 months post-randomisation	18 months post-randomisation
Weekly	Within clinic %SS (SR)	8.1 (6)	2 (3)	1.5 (3)	1.8 (2.5)
	Beyond clinic %SS (SR)	9.0 (5.5)	-	2.0 (2.3)	0.7 (1.5)
Twice Weekly	Within clinic %SS (SR)	6.6 (5)	1.4 (2)	1.5 (3)	0.3 (2)
	Beyond clinic %SS (SR)	10.3 (6.8)	-	2.4 (2.5)	1 (2)
Fortnightly	Within clinic %SS (SR)	9.0 (5)	1.1 (2)	1.1 (2.5)	0.7 (2)
	Beyond clinic %SS (SR)	7.9 (5)	-	1 (2)	1.4 (1.6)

Missed Sessions

To determine parental adherence to the Lidcombe Program schedule in the allocated treatment schedule, the average number of missed sessions was calculated for each group. This was calculated by counting the missed number of sessions for each participant and averaging the missed session data with other data from similar groups of children in this study. Data were available for all participants who completed Stage 1 and for participants up until the time of ceasing to attend the treatment schedule due to withdrawal or drop-out. Findings were available for 8 children in weekly, 9 in twice weekly and 12 in the fortnightly groups. The average missed sessions for the three groups was 4.4 in weekly, 8.7 in twice weekly and 1.0 in the fortnightly groups. The highest number of missed sessions was for children receiving twice weekly sessions and the lowest for children receiving fortnightly sessions.

Phone Consultations

The number of minutes spent in phone consultation was recorded on an Excel spreadsheet for each child. Although parents were encouraged to write down questions at home to discuss with the clinician at the scheduled clinic times, phone calls were accepted if the client needed. The total number of minutes of phone consultation for each group was 94 minutes in the weekly, 40 minutes in the twice weekly and 90 minutes in the fortnightly group. Clearly, the total minutes for the twice weekly group was the lowest, due to the higher frequency of visits and opportunity to ask the clinician questions on a more frequent basis, compared to the weekly and fortnightly groups.

Parent Questionnaire

The parents of 31 children recruited for the research were sent a questionnaire regarding their experience with the Lidcombe Program in their allocated treatment schedule. Parents were also asked to rate their satisfaction with their child's present

level of fluency on a scale of 1-10, where 1 = extremely satisfied and 10 = extremely dissatisfied. No questionnaires were returned from participants who had dropped out or were withdrawn from the research. Of the 21 children who completed the treatment in their allocated schedule, 16 parents (response rate of 76%) returned the questionnaire to the author. Of the 16 questionnaires, four were from the weekly group, six from twice weekly and six from the fortnightly group.

All 16 parents indicated that the Lidcombe Program was a positive experience. One parent comment was, "I liked the Lidcombe Program as I could see positive benefits and that there was a common goal that we were working towards." Another parent wrote, "I loved it because the program was positive and put the control and speed in the families hands." Another wrote, "The program allowed us to treat our son's stuttering with little to no disruptions to our daily routine and was flexible in terms of adapting to our son."

In the weekly group, all four parents (100%) indicated that the frequency of clinic visits was appropriate and that they would choose the same frequency if they were to start again. One parent from this group commented, "a higher frequency may have made it difficult to juggle and lesser frequency may have caused less compliance at home." On the scale of 1-10 for satisfaction with child's present level of fluency, three parents rated 1 and one parent rated 2 indicating that most parents were extremely satisfied.

In the twice weekly group, 3 of 6 parents (50%) indicated that the frequency of clinic visits was not appropriate due to the following reasons: 1) difficulty finding the time to attend twice weekly sessions, 2) far distance from home to clinic, and 3) insufficient time between visits for progress to occur. All three parents wrote that they would choose weekly sessions if they were to start again. Interestingly, one parent

commented, “twice weekly was helpful at the beginning for the first month but then it would have been good to move to once a week.” On a scale of 1-10 for satisfaction with child’s present level of fluency, two parents rated 1, three parents rated 2 and one parent rated 3, indicating that most parents were very satisfied with their child’s fluency.

In the fortnightly group, five out of six parents (83%) thought the frequency of clinic visits was appropriate and four parents indicated that they would choose the same frequency if they were to start again. Some comments from these parents were, “It would have been impossible for us to meet on a weekly basis so fortnightly was ideal.” Another wrote, “I was happy with the frequency and wouldn’t change it.” However, two parents indicated that weekly therapy would have been preferable, due to the frequency of clinical support and maintaining motivation. One parent commented that s/he would, “choose weekly for the first 1-2 months, then fortnightly after to gain more support and opportunity to ask more questions when treatment first begins and confidence is low.” Another parent commented that s/he would choose “once a week to keep things fresh but not excessive.” On a scale of 1-10 for satisfaction with child’s present level of fluency, four parents rated 1, one parent rated 2 and one parent rated 3, indicating that most parents were very satisfied with their child’s fluency.

A copy of the collated parent questionnaire responses is provided in Appendix Table A.3.

Discussion

In Chapter 4, a problem of clinical translation of the Lidcombe Program manual was discovered in that weekly treatment was not being achieved in clinical communities. To date no research has explored if scheduling Lidcombe Program clinic visits greater or less than weekly sessions is as efficient or efficacious as the standard. This study evaluated the effects of different treatment schedules on both the efficiency

and efficacy of the Lidcombe Program. A prospective Phase II RCT was conducted over two treatment sites in Australia and Canada. Thirty-one children were randomised into one of three treatment schedules: 8 in weekly sessions (the control group), 11 in fortnightly sessions (less intensive) and 12 in twice weekly sessions (more intensive), the latter two of which were the experimental groups. Overall, 21 children completed treatment in their allocated schedule, 7 in weekly, 6 in twice weekly and 8 in fortnightly sessions.

To determine the effect of different treatment schedules on the efficiency of the Lidcombe Program, primary and secondary outcomes were obtained. The primary outcome was the number of clinic visits and the secondary outcome was the number of weeks to complete Stage 1. The median numbers of clinic visits to complete Stage 1 for the treatment groups were 10 for fortnightly, 23 for standard weekly and 27 for twice weekly sessions. In comparison with worldwide benchmarking data (Koushik et al., 2011), children in the weekly group completed the first stage in approximately the 90th percentile clinic visits. A significant difference between treatment groups for the primary outcome clinic visits ($p = .01$) was found. No significant difference was found between the twice weekly and weekly treatment schedules ($p = .28$). However, fortnightly treatment was strongly associated ($p = .02$) with fewer clinic visits to complete Stage 1 compared to weekly treatment. In other words, children who attended fortnightly sessions reached near-zero levels of stuttering in fewer clinic visits than those in the other treatment schedules. The findings are comparable to results obtained from the file audit study of 134 children in North America general clinics presented in Chapter 4. Results from the file audit showed that children who attended the clinic less frequently (11 days or more apart) took fewer clinic visits to complete Stage 1 than those attending more frequently (fewer than 11 days apart).

For the secondary outcome measure, no difference was found between the treatment schedules for the number of weeks to complete Stage 1 (*logrank* $p = .748$). The median weeks by treatment schedule were 18 for fortnightly clinic visits, 23 for weekly and 16 for twice weekly. Children in the fortnightly group did not take longer to complete Stage 1 than those in the weekly or twice weekly groups. Therefore, less intensive treatment did not predict a longer duration of treatment. Similarly, children in the twice weekly group did not complete Stage 1 more quickly than children in the other groups. Therefore, intensive sessions did not predict a shorter duration of treatment. This is an important finding from a service delivery perspective. If replication with larger numbers of children shows similar results, fortnightly sessions might be a more efficient model in treating early stuttering with fewer clinic visits in the same number of weeks, compared to weekly or twice weekly clinic visits.

The effect of different treatment schedules on the efficacy of the Lidcombe Program was determined by comparing the stuttering severity on different assessment occasions to previous efficacy research (Jones et al., 2005). Stuttering severity was determined by two speech measures, %SS and SR. When the data from the different treatment schedules were combined, the median beyond clinic %SS (SR) measures for all children was 8.2 (5.5) pre-treatment, 1.9 (2) at 9 months post-randomisation and 1.2 (2) at 18 months post-randomisation. It is noted that the number of samples collected at 18 months post-randomisation was less than 50% than the required amount, which is a limitation of this study. However, the collected samples at 9 months post-randomisation were adequate (83%). The results of the present study were similar to those reported by Jones et al. In a Phase III RCT of the Lidcombe Program, Jones et al. found the mean %SS for 29 children at 9 months post-randomisation to be 1.5. By treatment schedule, the median %SS at 9 months post-randomisation was 2.4 for twice weekly, 1.0 for

fortnightly and 2.0 for weekly. In comparison to the Jones et al. 9-month post-randomisation data, the treatment schedules did not affect the efficacy of treatment outcomes for children in the fortnightly group. Thus, for the children in this study, the less frequent fortnightly sessions appeared as efficacious as the standard Lidcombe Program.

The results of the parent questionnaire showed that 83% of the families in the fortnightly group were happy with their allocated treatment schedule and would choose the same schedule if they were to start over. The average number of missed sessions was lowest for the fortnightly group in comparison to the weekly and twice weekly schedules. This might be due to the fact that motivation to meet treatment demands on this schedule was high. If a treatment was missed on a fortnightly schedule the clinician could re-schedule another appointment for the same week, as per the protocol. However if the client was unavailable, the next available clinic visit would be a fortnight later, almost one month since the child's last clinic visit. In the twice weekly group, half of the families found the intensive schedule difficult to maintain due to demands at home, work, and travel time to the clinic. If a twice weekly session was missed, the family would still have the opportunity to see the clinician another day that same week, or if not shortly the week after, resulting in minimal time gaps between clinic visits. Thus, the motivation to maintain twice weekly schedules might not have been as high for the families attending less frequently. This conjecture is supported by the fact that the average number of missed sessions for this group was the highest of all three treatment schedules. Finally, in the weekly group all families were happy with their treatment schedule and would choose the same frequency of visits if they were to start again. However, the average number of missed sessions was higher than that for the fortnightly group. This might be due to the fact that a missed session could be rescheduled later on

that week, as per the protocol, or the child would be seen by the clinician only 2 weeks after the last clinic visit.

Of all participants who completed treatment in their allocated schedule, over half (56%) of the families indicated that they would choose weekly treatment if they were to start over. Thus, the remainder of families were willing to consider an alternative service delivery model. Most families in the fortnightly group (83%) reported that they enjoyed the frequency of clinic visits. This group had the fewest average number of missed sessions as compared to the other treatment schedules. Furthermore, except for one client who dropped out due to moving cities, the remainder of the fortnightly participants completed Stage 1 of the program. The three who were removed from the fortnightly group were removed due to the protocol rather than wanting to change to weekly treatment.

Although twice weekly treatment was efficacious in treating stuttering, it was not more efficient than weekly or fortnightly treatment. The median number of clinic visits to complete Stage 1 was more than double that for fortnightly visits. Further, the clinician time was doubled to 2 hours a week for each client; the number of drop-outs was highest, and the participants in this group had the highest average number of missed sessions. Due to the protocol restrictions, two children were removed from this group and offered weekly treatment. The families did not continue with treatment, possibly suffering burnout. Clearly, the twice weekly group was the least preferable of all three service delivery groups.

Although this preliminary study was based on small number, the result has implications for service delivery models with the Lidcombe Program. Fortnightly sessions might be a more appropriate alternative to weekly treatment. The evidence presented in this empirical study contributes to the external evidence in EBP. With

replication, similar findings can benefit clinical communities. As waiting lists are increasing around the world, health institutions might offer children an alternative to the standard weekly sessions. By this means, more children could be treated for stuttering by a speech pathologist in alternating weekly time slots. This would benefit waiting lists by reducing times for treatment. Further, clinicians and parents could better manage their schedules, as clinic visits could be appropriately scheduled. If replication and extension produce similar results, the findings could greatly improve service delivery options of the Lidcombe Program.

Summary

The review of treatment process evidence of the Lidcombe Program in Chapter 3 found a gap in the literature with regards to the regression studies in Australia and the United Kingdom (Jones et al., 2000; Kingston et al., 2003). Predictor variables were explored. However, the average time between clinic visits was not assessed. In clinical communities, adhering to weekly Lidcombe Program treatment appeared difficult due to policy, parent, child or clinician factors. Further evidence presented in Chapter 4 found that weekly Lidcombe Program treatment was not being achieved in clinics. Clearly, a clinical translation problem of the Lidcombe Program manual was found, thus prompting the prospective study in this chapter. The present study contributes to external systematic evidence in EBP. As this area of research has not been explored in previous studies, this is the first prospective study. The findings from this study provide important preliminary evidence on Lidcombe Program treatment schedules.

The following chapter presents a discussion of key findings from the two empirical studies, implications for clinical practice, theoretical implications, limitations and concluding remarks.

Chapter 6

Treatment Schedules of the Lidcombe Program

The primary purpose of this thesis was to explore clinical translation and benchmark research of the Lidcombe Program and to extend findings in a prospective study to improve clinical trials research. This was achieved by two empirical studies designed first to evaluate treatment schedules in real-world clinical communities and second in controlled laboratory contexts.

The first empirical research evaluated predictor variables and the relationship with the number of clinic visits to complete Stage 1 of the Lidcombe Program. This was achieved by a retrospective file audit in North-American general clinics. The data obtained from the file audit were combined with data from previous studies in Australia and the United Kingdom to determine worldwide clinical benchmarks.

The results of the North American file audit informed and led to the development of the Phase II clinical trial, the second empirical research of this thesis. The clinical trial was a carefully designed prospective study varying the treatment schedules of the Lidcombe Program and evaluating the effects on treatment efficacy and efficiency. The purpose was to determine whether altering the frequency of Lidcombe Program clinic visits could be supported by evidence. The key findings of both empirical studies are presented in the following section.

Key Findings

The Lidcombe Program in North America

The North American file audit investigated the effects of the Lidcombe Program in general clinics by evaluating the median and 90th percentile number of clinic visits for children to complete Stage 1. The median number of clinic visits in which 50% of the cohort had completed Stage 1 was 12. In the Australian and British studies, the

median number of clinic visits was similar, at 11 visits for both studies. Further, 90% of all North American children completed Stage 1 in 22 visits. Similarly in the Australian and British studies, the 90th and 95th percentiles were 22 and 21 clinic visits respectively. All three studies independently agreed on the percentile values. The results showed that children treated with the Lidcombe Program in North America obtained benchmarks similar to those obtained by children in Australia and the United Kingdom.

World-wide Benchmarks for Delivery of the Lidcombe Program

World-wide benchmarks were obtained by combining data from the most recent file audit and the Australian and British studies. The pooled data of 444 children was substantial, thus increasing the statistical power. Meta-analysis was applied for the purpose of providing clinical benchmarks. Findings showed that 444 children required a median of 11 clinic visits to reach near-zero levels of stuttering. Further, 90% of the children required 22 clinic visits. These benchmarks can serve as a guide for clinicians in their own clinical practice.

Predictors of Treatment Time in the Real World

Age, Gender and Onset-to-Treatment Interval

No evidence was found in the North American data of an association between the number of clinic visits and the predictor variables age, gender, or onset-to-treatment interval. As in the Australian and British studies, no evidence of an association with the number of clinic visits was found for the predictor variables age and gender. Contrary to the Australian and United Kingdom studies, however, the North-American file audit found no association between onset-to-treatment interval and number of clinic visits during Stage 1.

Severity

The North American file audit found strong evidence that higher pre-treatment severity (5.0% or more) was associated with an increased number of Stage 1 clinic visits, as was found in the Australian and British studies. In the meta-analysis of 444 case files, severity was re-categorised into three levels. The median number of clinic visits required to complete Stage 1 was 10 for mild (0-4.9%SS), 12 for moderate (5.0-9.9%SS), and 14 (10%SS +) for severe pre-treatment severity of stuttering.

Average Time between Clinic Visits

The average time between clinic visits for the predictor variable frequency of attendance was found to be 11 days. This study thus provided evidence that Lidcombe Program weekly visits were not being achieved. There was some evidence that frequent clinic attendance was associated with a higher number of Stage 1 clinic visits. In other words, children who attended the clinic more often than every 11 days required more clinic visits than those who attended less frequently. This was a surprising finding; the expectation had been that increased frequency of visits might predict fewer Stage 1 clinic visits. This information added to the evidence base regarding a new predictor variable which had not been previously explored. The file audit provided important information on attendance frequency in general clinics, but still did not inform us of the optimal treatment schedules of the Lidcombe Program.

Efficacy and Efficiency of the Lidcombe Program

Treatment Schedules

The Phase II clinical trial found that a fortnightly treatment schedule was strongly associated with fewer clinic visits than for twice weekly and weekly treatment. In other words, children who attended on a fortnightly basis reached near-zero stuttering in fewer clinic visits than those in the other treatment schedules. The median number of

clinic visits required to complete Stage 1 by treatment schedule was 27 for twice weekly, 23 for weekly and 10 for fortnightly sessions. Compared to worldwide benchmarks established in Chapter 4, children in the weekly group did not complete Stage 1 in the median treatment time of 11 clinic visits. This may be due to the small number of children in the group (N=7). Further, one child required 40 clinic visits to complete the first stage due to behavioral issues that required family psychological counseling. However, children in the weekly group did complete Stage 1 in the 90th percentile clinic visits as compared to worldwide benchmarks.

The effect size showed that children in the fortnightly group were 3.8 times more likely to have fewer clinic visits than children in the weekly group. This was similar to the results established in the North American file audit, in which children attending the clinic less frequently, with 11 days or more between sessions, required fewer clinic visits than those attending more frequently, with fewer than 11 days between sessions. Thus the findings of this study were that fortnightly sessions were more efficient than weekly or twice weekly treatment.

No significant differences were found between weekly, twice weekly and fortnightly treatment schedules for number of weeks to complete Stage 1. The median number of weeks by treatment schedule was 23 for weekly, 16 for twice weekly and 18 for fortnightly sessions. In other words, all the children in this study completed Stage 1 in approximately the same number of weeks, regardless of treatment schedule.

Stuttering Severity

Stuttering severity was rated in percent syllables stuttered (%SS) and severity ratings (SR) by a blinded observer at pre-treatment, 9 months post-randomisation and 18 months post-randomisation, for all BC samples. The data were compared to 9 months post-randomisation data from a Phase III RCT of the Lidcombe Program (Jones

et al., 2005). The medians for all children grouped together in beyond clinic %SS (and SR) were 7.9 (5.5) pre-treatment, 1.9 (2) at 9 months post-randomisation and 1.2 (2) at 18 months post-randomisation. Findings were similar in comparison with Jones et al. (2005) with a mean %SS at 9 months post-randomisation at 1.5. By treatment schedule, the median %SS (and SR) at 9 months post-randomisation was 2.4 (2.5) for twice weekly, 2.0 (2.3) for weekly and 1.0 (2.0) for fortnightly sessions. The fortnightly treatment schedule produced similar findings to the Phase III clinical trial by Jones et al., thus the efficacy of the Lidcombe Program was not affected by the treatment schedule.

Parent Questionnaire

The parent questionnaire revealed that all participants enjoyed the Lidcombe Program. Over half of the families indicated that they would choose weekly treatment if they were to start over. However, the remainder of the families were willing to choose an alternative treatment schedule to suit their schedule. In the fortnightly group, 83% enjoyed the frequency of clinic visits. However, only 50% enjoyed the schedule in the twice weekly group. This finding was supported by the average number of missed sessions in each group. By treatment schedule, the average numbers of missed sessions were 4.4 in the weekly, 8.7 in twice weekly and 1 in the fortnightly groups. The fortnightly group had the lowest average number of missed sessions and the twice weekly group had the highest, most of which were due to client cancellations.

Participant Attrition

Dollaghan (2007) described attrition as when there are fewer participants at the end of the study than the beginning. Attrition can affect the external validity of a study, and therefore full disclosure regarding withdrawal from research is important. In this study, 10 (32%) participants were withdrawn by the research protocol or independently

dropped out of the research. Of these children, four (40%) were withdrawn due to the protocol of the study. The ethical arrangements for this study required that children still receiving treatment in the experimental groups after 6 months' duration were to be removed and placed in the weekly group. This was problematic for the fortnightly group, as the number of clinic visits at 6 months approached the median number of clinic visits required for 50% of children to complete Stage 1 (Jones et al., 2000). This oversight in protocol planning meant that two children from the fortnightly group were required to move into the weekly group even though treatment gains were being achieved. Of the children who independently dropped out of research, three (30%) families could not keep up with the twice weekly schedule. The twice weekly treatment schedule was found to be the least preferred, as the majority of participants who independently left the research program were from that group.

Clinical Implications

Findings from the two empirical studies had implications for clinical practice and EBP. The results of the empirical studies contributed to systematic research evidence in EBP.

The worldwide benchmarks established from combining data from three English speaking countries in a meta-analysis provided data for clinical translation. Although clinical benchmarks had been available from two countries, Australia and the United Kingdom (Jones et al., 2000; Kingston et al., 2003), the addition of the North American data strengthened the statistical findings. As EBP becomes increasingly essential, the established benchmarks have provided clinicians with standards for treatment time with the Lidcombe Program. The findings were based on real-world situations in clinical communities rather than controlled research contexts. Therefore, the benchmarks took into account service providers' policies regarding treatment schedules, and cancellations

that occurred due to parent, child or clinician factors. The worldwide benchmarks for median and 90th percentile number of clinic visits required to complete Stage 1 were 11 and 22 respectively. With these benchmarks, clinicians can monitor a child's progress with the Lidcombe Program by comparing with the standard. If treatment time during Stage 1 is extended beyond the benchmarks, consultation or assistance with such cases should be immediately sought.

The meta-analysis re-classified stuttering severity into mild, moderate and severe, increasing the specificity of findings for the median number of clinic visits required to complete Stage 1. Further, the new classification of mild, moderate and severe is familiar to speech pathologists, who refer to other speech and language disorders in similar categories. Results of the meta-analysis showed that the median number of clinic visits required to complete Stage 1 were 10 for mild (1-5%SS), 12 for moderate (6-10%SS), and 14 (11%SS or more) for severe pre-treatment stuttering. That information is important for clinicians planning treatment time for individual clients. If a child presents with severe stuttering of 11%SS or more, the clinician can expect that the median number of clinic visits would be extended to approximately four more sessions than for a child presenting with mild stuttering. Therefore, pre-treatment severity can be a predictor for the number of clinic visits required during Stage 1. Parents can be informed of expected treatment time based on their child's severity of stuttering and thus can make appropriate plans. Further, funding bodies and health care systems can make informed decisions regarding allocation of monies and number of allocated clinic visits for a child waiting for early stuttering treatment with the Lidcombe Program.

The average time between clinic visits in the North American file audit was 11 days, rather than the suggested 7 days in the manual (Packman et al., 2010). This was

similar to the results found by O'Brian et al. (2011), who determined the average time between clinic visits as 15 days, again more than 7 days. Both studies suggested that weekly treatment was not being achieved in clinical communities. However, the data from the North American file audit further suggested that clinic visits more than 7 days apart might be more efficient than the standard. A longer interval between clinic visits (average 11 days or more) resulted in fewer clinic visits than a shorter time between clinic visits (average less than 11 days).

In the Phase II clinical trial, strong evidence was found for the association of fortnightly clinic visits and treatment time during Stage 1. Fortnightly sessions were superior to the weekly and twice weekly treatment schedules. The median number of clinic visits was reduced, drop-outs were infrequent, and the average number of missed sessions was lower than for the other groups. Further, the efficacy of the Lidcombe Program was not compromised by fortnightly treatment. The results at 9 months post-randomisation were comparable to Phase III outcomes research (Jones et al., 2005). The implications of these findings are vast. As highlighted in Chapter 5, waiting lists for treatment services are problematic (O'Leary, 2010). Fortnightly treatment would benefit waiting lists as more children could be seen in alternate time slots. This would reduce both the wait times for children requiring stuttering treatment and the overall number of children waiting for services. Parents could manage their schedules more easily, as clinic visits would occur bi-monthly instead of four times per month. Funding bodies could allocate more monies to children requiring stuttering treatment with the Lidcombe Program, as twice as many children could be seen in fewer clinic visits.

Theoretical Implications

The theoretical issues discussed in this section pertain to the efficiency of Lidcombe Program treatment schedules. Surprisingly, the median number of clinic

visits for children receiving fortnightly sessions was lower than those for the weekly and twice weekly schedules. In other words, a less frequent schedule resulted in faster treatment time, whereas the expectation might be the opposite.

The finding may be attributable to the time required for response contingent stimulation procedures to demonstrate changes in stuttering frequency. Children in the fortnightly group had more time between clinic visits than those in the twice weekly and weekly groups. Changes in speech motor processing may require time. Children in the twice weekly group might have had insufficient time between clinic visits to regain speech motor control, thus requiring more clinic visits.

Parent factors might also be related to the research findings. The infrequency of fortnightly clinic visits might have prompted parents to be proactive in the treatment process rather than be reliant on the clinician. Parents in the fortnightly group might have taken more of a lead in responding to their child's stuttering. This could have resulted in greater adherence to daily treatment goals, with parents being active participants in therapy sessions and asking relevant questions. On the other hand, parents in the twice weekly group might have relied more on the therapist to guide the treatment process, might have been less proactive and might have adhered less to consistent daily treatment goals, because the child was seen frequently for treatment. Parents in the fortnightly group might have developed better troubleshooting skills whereas those in the twice weekly group might have relied on the clinician to solve problems, thus resulting in faster results in the fortnightly group.

Limitations

The North American file audit was retrospective in nature. A limitation of retrospective data is that the researcher depends on the accuracy and availability of the data. Missing data or inaccuracy of reporting the variables of interest is possible in such

designs. Another limitation is that retrospective studies are prone to selection bias because the researcher selects the cases. Finally, retrospective studies are uncontrolled in that there is no comparison to a control group.

The Phase II clinical trial was based on small subject numbers, and therefore results can be attributed to the participants in that study only. Further, at the time of analyses of data, the collected samples for the 18 month post-randomisation data was less than 50% of the total group. Thus, the 18 month total group findings for %SS and SR are not representative of the total group. The collected samples for the 18 month data for the weekly and twice weekly group were also less than 50% and therefore were not representative for those groups. However, the results for the fortnightly group were based on adequate representation of that group. The significant associations reported merit larger clinical trials to measure the effects with a higher number of participants.

Future Directions

Phase III Clinical Trial

The Phase II clinical trial, as mentioned, was based on small subject numbers and requires replication with larger numbers of children. However, the findings reported in this study for the twice weekly schedule indicated that the schedule was more time consuming, had the highest drop-out rates, the highest number of missed sessions, and was not more efficient in terms of number of weeks than the other treatment schedules. It is recommended, therefore, that any replication of this methodology should omit the twice weekly schedule as it was found to be the least efficient of the three groups. Replication should compare weekly with fortnightly treatment schedules, with larger numbers of subjects, to corroborate the findings from this study.

Alternative Treatment Schedules

A treatment schedule based on an alternative model, such as weekly clinic visits for 4 weeks followed by fortnightly treatment until the completion of Stage 1 might be a direction for future research. In the parent questionnaire, one parent wrote that she would choose weekly treatment for 1-2 months and fortnightly thereafter, to gain confidence during the early weeks. This suggestion was supported by the fact that two parents in the fortnightly group initiated phone consultations with the clinician after only receiving one treatment session. Research indicates that stuttering severity in the Lidcombe Program should reduce by one-third, 4 weeks after the start of treatment (Onslow & Yaruss, 2007). At this time, parents might have more confidence in the program requirements and thus feel ready to move to fortnightly sessions after the initial four weekly clinic visits. This may be an area for further exploration.

Fortnightly Treatment for School-Age Children

The Lidcombe Program has been shown to be effective for school-age children (Koushik et al., 2009; Lincoln et al., 1996). School-age children are increasingly busy, with requirements for school and social activities becoming priorities. It is recommended that a similar methodology for alternative treatment schedules be trialled with school-age children. The trial could include weekly and fortnightly schedules to determine the effects of the Lidcombe Program on number of clinic visits for this population. If fortnightly sessions are found to be efficacious with school-age children, the required time for clinic visits could be less strenuous for the family with bi-monthly rather than weekly sessions.

Concluding Remarks

The findings established from this research suggest that fortnightly sessions might be an appropriate service delivery model for treatment of early stuttering with the

Lidcombe Program. The treatment schedule was found to be both efficient and efficacious for the children in this study. Less frequent clinic visits would benefit health care systems, funding bodies, clinicians and parents. It is hoped that the findings from this research will prompt larger scale investigations to confirm the viability of fortnightly sessions as an alternative service delivery model for the Lidcombe Program.

Investigations of other treatment schedules might provide another alternative model to benefit waiting lists. Further, fortnightly sessions might benefit school-age children as the curriculum and social demands increase during those years. It is hoped that the research findings from this thesis will encourage further studies. These areas are possibilities for further inquiry to add to the evidence base of an efficacious treatment for stuttering.

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APPENDICES

Table A.1: Details of randomised participants: gender, age, family history, speech and language delay, treatment schedule

<i>Participant</i>	<i>Gender</i>	<i>Age at Assessment (months)</i>	<i>Family History of Stuttering</i>	<i>Speech and Language Assessment Results</i>	<i>Randomly Assigned Treatment Schedule</i>
1	Male	67	No	Moderate Language Delay	Weekly
2	Male	36	No	-	Weekly
3	Male	53	Yes	Mild Language Delay	Weekly
4	Male	44	No	Mild Language Delay	Weekly
5	Male	36	Yes	-	Weekly
6	Male	58	Yes	-	Weekly
7	Male	48	No	-	Weekly
8	Male	45	No	-	Weekly
9	Male	50	Yes	-	Twice Weekly
10	Male	54	No	Mild Language Delay	Twice Weekly
11	Male	56	Yes	Moderate Phonological & Mild-Moderate Language Delay	Twice Weekly
12	Female	42	No	Mild Language Delay	Twice Weekly
13	Male	52	Yes	-	Twice Weekly
14	Male	65	No	-	Twice Weekly
15	Male	47	Yes	-	Twice Weekly
16	Male	47	No	-	Twice Weekly
17	Male	47	No	-	Twice Weekly
18	Female	55	No	Mild Language Delay	Twice Weekly
19	Female	46	Yes	-	Twice Weekly
20	Female	50	Yes	-	Fortnightly
21	Male	51	No	Mild Language Delay	Fortnightly
22	Male	51	No	Moderate Phonological Delay	Fortnightly
23	Male	62	No	Mild Language Delay	Fortnightly
24	Male	57	Yes	-	Fortnightly
25	Male	59	Yes	Mild-Moderate Language Delay	Fortnightly
26	Female	60	Yes	-	Fortnightly

27	Male	40	Yes	-	Fortnightly
28	Male	38	No	-	Fortnightly
29	Male	44	No	Mild Language Delay	Fortnightly
30	Male	40	Yes	-	Fortnightly
31	Male	54	Yes	-	Fortnightly

Table A.2: Primary and secondary outcomes and BC %SS and SRs at each assessment occasion

<i>Participant Number</i>	<i>Group</i>	<i>Clinic Visits</i>	<i>Weeks</i>	<i>Pre-treatment %SS and (SR)</i>	<i>9-months %SS and (SR)</i>	<i>18-months %SS and (SR)</i>
1	Weekly	12	11	7.1 (5)	3.5 (4)	1.4 (2)
2	Weekly	23	23	12.6 (7)	6 (5)	0 (1)
3	Weekly	40	46	9.5 (5.5)	0.9 (2)	
4	Weekly	39	33	8.5 (5.5)	2.1 (2)	
5	Weekly	15	17	4.3 (3)	0.8 (2)	
6	Weekly	11	15	5.4 (4.5)		
7	Weekly	28	30	10.5 (6.5)	1.9 (2.5)	1.2 (2)
8	Twice Weekly	11	7	2.8 (2.5)	2.4 (2.5)	
9	Twice Weekly	10	9	13 (6.5)	0.1 (1.5)	1.0 (2)
10	Twice Weekly	27	16	17.3 (7)	2.5 (3)	0 (1)
11	Twice Weekly	27	16	14 (7)	0.5 (2)	
12	Twice Weekly	29	20	5.8 (5)	3 (4.5)	
13	Twice Weekly	47	33	7.6 (7)	0.6 (2)	
14	Fortnightly	6	10	7.2 (5)	0 (1)	1 (1)
15	Fortnightly	9	16	5.9 (5.5)	1.9 (2)	
16	Fortnightly	13	24	8.9 (5)	1.3 (2.5)	1.3 (1.2)
17	Fortnightly	17	34	2.2 (3)	0.3 (2)	1.4 (2)
18	Fortnightly	6	10	2.4 (2.5)	2 (2)	0 (1)
19	Fortnightly	10	20	11.5 (7)	0.7 (2)	
20	Fortnightly	7	15	3.4 (4.5)	3.3 (3.5)	3.1 (5)
21	Fortnightly	23	46	14.9 (7)	0.6 (2)	
22	Fortnightly	20	49	7.9 (5)		
23	Fortnightly	30	60	15.2 (7)		3.4 (4)
24	Fortnightly	66	100	9.7 (6.5)		

Table A.3: Parent questionnaire responses

Parent Question	Weekly (N=4)	Twice Weekly (N=6)	Fortnightly (N=6)
1) On a scale of 1-10, where 1= no stuttering and 10= extremely severe stuttering, what is the typical average severity rating of your child's speech during the last week?	1 = 3 parents 2 = 1 parent	1 = 4 parents 2 = 2 parents	1 = 4 parents 2 = 1 parent 3 = 1 parent
2) On a scale of 1-10, where 1= no stuttering and 10= extremely severe stuttering, what is the most severe severity rating of your child's speech during the last week?	1= 3 parents 2 = 1 parent	1 = 2 parents 2 = 4 parents	1 = 4 parents 3 = 1 parent 4 = 1 parent
3) On a scale of 1-10, where 1=extremely satisfied and 10=extremely dissatisfied, how satisfied are you with your child's present fluency?	1= 3 parents 2= 1 parent	1= 2 parents 2= 3 parents 3= 1 parent	1= 4 parents 2= 1 parent 3= 1 parent
4) Did you like the Lidcombe Program?	4 parents = Yes	6 parents = Yes	6 parents = Yes
5) What did you like about the Lidcombe Program?	-It has improved his stutter -Love the flexible program -Loved the program, liked being involved	-Child-clinician rapport -Simple exercises -Very effective -Works wonders -Easy to understand and participant in -Evidence to support program -My child can be understood - Loved the program, it taught me what to do for my son's stutter	-Positive benefits with the LP -Helpful program for stuttering treatment -My son is now more confident -Found program simple and effective
6) Did you have enough clinical support?	4 parents = Yes	6 parents = Yes	6 parents = Yes
7) Was there enough progress between clinic visits?	3 parents = Yes 1 parent = No	5 parents = Yes 1 parent = No	5 parents = Yes 1 parent = No

8) If not, why not?	-Thought he would finish quicker	-Not enough time between visits -Progress slow when I hadn't worked much at home	-Too slow
9) Did you have enough confidence to carry out treatment activities at home?	4 parents = YES	6 parents = Yes	6 parents = Yes
10) Do you think that the frequency of clinic visits was appropriate?	100% Yes	50% Yes 50% No	83% Yes 17% No
11) Why or why not?		- difficult to attend due to work/ school/ needs of other siblings - difficult to attend due to time/distance from clinic - Not enough time between visits for progress to occur	- I think my child would have been a bit more engaged if we had gone once a week because by the end of the second week we were both losing steam, but then a next visit always got us excited again
12) If you were start over and choose an ideal frequency of clinic visits, what would you choose?	4 parents = Weekly	3 parents = Twice Weekly 3 parents = Weekly	4 parents = Fortnightly 2 parents = Weekly
13) Was there anything challenging?	4 parents = No	6 parents = No	6 parents = No
14) Other comments	- Higher frequency may have made it difficult to juggle; lesser frequency may have caused less compliance at home	- Twice weekly was helpful at the beginning (maybe for 1 st month) but then would have been good to move to once a week -For me, I could take the time to go twice a week since I was working from home	-It would have been impossible for us to meet on a weekly basis, so fortnightly was ideal - I would choose weekly for the first 1-2 months, then fortnightly after to gain more support and opportunity to ask more questions when treatment first begins and confidence is low -Once a week to keep things fresh but not excessive