

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

March 2012

STATEMENT OF ORIGINALITY

This thesis contains no material which has been accepted for the award of any other degree or diploma in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. I give consent to this copy of my thesis, when deposited in the University Library**, being made available for loan and photocopying subject to the provisions of the Copyright Act 1968.

****Unless an embargo has been approved for a determined period.**

Sarita Koushik, PhD Candidate

ACKNOWLEDGEMENT OF AUTHORSHIP

I hereby certify that the work embodied in this thesis contains a published paper/s/scholarly work of which I am a joint author. I have included as part of the thesis a written statement, endorsed by my supervisor, attesting to my contribution to the joint publication/s/scholarly work.

Endorsement by Supervisor:

Sally Hewat, PhD

ACKNOWLEDGEMENTS

This thesis would not have been possible without the guidance and support from my supervisors, family and friends.

First, I owe my deepest gratitude to my primary supervisor, Dr Sally Hewat for her strong mentorship from the start to the end of this thesis. I especially acknowledge her kind words during times of struggle and advice to focus on the light at the end of the tunnel. I am grateful to Professor Mark Onslow for giving me the opportunity to begin this journey and for his thorough mentorship over the years. I would like to thank Dr Rosalee Shenker for her mentorship, for providing funding and a facility for research at the Montreal Fluency Centre. Particular thanks are extended to Alison Ferguson for her knowledge, support and positive energy. I am honoured to have worked with such knowledgeable researchers and am grateful for the entire experience.

My deepest thanks are extended to colleagues Danra Kazenski, Julie McFarlane and Monica Anderson for their clinical work on this project and for treating each child with the greatest care. Huge amounts of thanks are extended to Vanessa Harris for her time and effort rating all speech samples for this project. Many thanks are extended to Joanne Wilding for proofreading and editing the first thesis draft on her own free time.

I would like to express my sincere gratitude to the parents and children who participated in this research project. Without them, the research achieved in this thesis would not have been possible. I am extremely grateful for the scholarships I received from the National Health and Medical Research Council (NHMRC) and University of Newcastle International Scholarship Program.

My loving thanks are extended to my mom and sister. The support they provided throughout the years was invaluable and I feel grateful to have been blessed with a

wonderful family. To my friends, thank you for encouraging me to keep my chin up throughout this journey.

Finally, I would like to deeply and lovingly thank my partner Chris. Thank you for your kind words, love and support. Thanks for putting up with me and giving me your positive energy every single day. You have made all of my dreams come true and I truly could not have reached this milestone without you.

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

JOURNAL ARTICLES

Koushik, S., Hewat, S., Shenker, R. C., Jones, M., & Onslow, M. (2011). North-American Lidcombe Program file audit: Replication and meta-analysis. *International Journal of Speech Language Pathology*, 13(4), 301-307.

INTERNATIONAL CONFERENCE PRESENTATIONS

Koushik, S., Hewat, S., Shenker, R. C., Jones, M., & Onslow, M. (2010, November). The effects of varying different treatment schedules on Lidcombe Program clinic visits: A phase II clinical trial. Symposium conducted at the American Speech and Hearing Association Convention, Philadelphia, USA.

Koushik, S., Hewat, S., Shenker, R. C., Jones, M., & Onslow, M. (2009, August). North-American Lidcombe Program file audit: Replication and meta-analysis. Symposium conducted at the 6th International Fluency Association World Congress, Rio de Janeiro, Brazil.

POSTERS

Koushik, S., Hewat, S., Shenker, R. C., Jones, M., & Onslow, M. (2010 November). The effects of varying different treatment schedules on Lidcombe Program clinic visits: A phase II clinical trial. Symposium conducted at the University of Newcastle RHD Symposium, Newcastle, Australia.

TABLE OF CONTENTS

Publications, Presentations and Posters	vii
List of Tables	xi
List of Figures	xii
List of Acronyms	xiii
Précis	xiv
Abstract	xvi
 CHAPTER 1: Overview of Stuttering and Evidence-Based Practice	 1
Description of Stuttering	1
Cause of Stuttering	2
Development of Stuttering	2
Natural Recovery	4
Treatment for Stuttering	4
Impact of Stuttering	5
Evaluating Early Stuttering Treatments	5
Evidence Based Practice	6
External Evidence	6
Internal Evidence	9
Client Preference	10
Summary of Evidence Based Practice	10
Robey's Five-Phase Model	10
Evaluation of a Clinical Trial	12
Phases of a Clinical Trial	15
Summary	16
 CHAPTER 2: Evaluating Treatments for Early Stuttering	 18
Multi-factorial Treatments	19
Group Play Therapy	19
Demands and Capacities Model	20
Mother-Child Interaction Therapy	20
Parent-Child Interaction	21
Speech Restructuring	23
The Comprehensive Stuttering Program	23
The Fluency Rules Program	24
The Preschool Fluency Development Program	25
Intensive Stuttering Therapy Program	25
Syllable-Times Speech	26
Verbal Response Contingent Stimulation	27
Early VRCS Treatments	28
The Lidcombe Program of Early Stuttering Intervention	29
Phase I Clinical Trials	30
Phase II Clinical Trials	31
Phase III Clinical Trials	34
Summary of Early Intervention Approaches	35
Summary	37

CHAPTER 3: The Lidcombe Program of Early Stuttering Intervention	38
Rationale	38
Principles	39
Lidcombe Program Components	39
Parental Verbal Contingences	39
Measurement	40
Weekly Clinic Visits	40
Treatment in Structured and Unstructured Conversations	40
Primary Goals	41
Treatment Process Research	42
Natural Recovery	42
Long-Term Impact	43
Psychological Impact	43
Social Validity	44
Child and Parent Speech and Language	45
Cultural Impact	45
Implementation with School-Age Children	46
Parent Perception	47
Predictors of Treatment Time	49
Australian Study	49
United Kingdom Study	50
Meta-analysis	50
Summary	51
 CHAPTER 4: Lidcombe Program Outcomes in the Real World	 53
Method	54
Study Design	54
Ethics	54
Participants	55
Variables	56
Dependent Variable	56
Predictor Variables	56
Results	57
Median Number of Clinic Visits by Clinic Site	58
Median Number of Clinic Visits for the Group	59
Logistic Regression	60
Goodness-of-Fit	61
Meta-analysis	62
Discussion	63
 CHAPTER 5: Does changing the Frequency of Lidcombe Program Clinic Visits Affect Outcomes?	 67
Method	70
Study Design	70
Ethics	70
Research Sites	71
Randomisation	71
Inclusion and Exclusion Criteria	72
Treatment	73
Participants	74

Drop-outs and Withdrawal	75
Participants Who Completed Stage 1	77
Outcomes	78
Efficiency of the Treatment Schedules	79
Efficacy of the Treatment Schedules	79
Stuttering Severity	80
Inter-Judge and Intra-Judge Reliability	82
Parent-Questionnaire	82
Results	83
Efficiency of Treatment Schedules	83
Primary Outcome	83
Secondary Outcome	85
Efficacy of Treatment Schedules	86
Stuttering Severity	86
Missed Sessions	88
Phone Consultations	88
Parent-Questionnaire	88
Discussion	90
Summary	95
 CHAPTER 6: Treatment Schedules of the Lidcombe Program	 96
Key Findings	96
The Lidcombe Program in North America	96
World-wide Benchmarks for Delivery of the Lidcombe Program	97
Predictors of Treatment Time in the Real World	97
Age, Gender and Onset-to-Treatment Interval	97
Severity	98
Average Time between Clinic Visits	98
Efficacy and Efficiency of the Lidcombe Program	98
Treatment Schedules	98
Stuttering Severity	99
Parent Questionnaire	100
Participant Attrition	100
Clinical Implications	101
Theoretical Implications	103
Limitations	104
Future Directions	105
Phase III Clinical Trial	105
Alternative Treatment Schedules	106
Fortnightly Treatment for School-Age Children	106
Concluding Remarks	106
References	108
Appendices	120

LIST OF TABLES

Table 1.1. Levels of evidence hierarchy by NHMRC (2009) guidelines	7
Table 1.2. Robey's (2004) five-phase model and description	12
Table 1.3. Onslow et al. (2008) clinical trial phases	15
Table 2.1. Multi-factorial, speech restructuring and verbal response contingent stimulation models of early stuttering treatment and current level of evidence	35
Table 4.1. Descriptive statistics for the North American data (N=134)	58
Table 4.2. Results of univariable logistic regression for the North American data	61
Table 4.3. Results of the univariable logistic regression (Australian, British and North American cohorts)	62
Table 5.1. Summary of recruited participants by group	75
Table 5.2. Details of participant drop-out or withdrawal	77
Table 5.3. Details of children who completed Stage 1 in treatment schedule	78
Table 5.4. Number of within- and beyond-clinic recordings on each assessment occasion	80
Table 5.5. The total number of collected WC and BC speech samples on each assessment occasion	81
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.	87
Appendix Table A.1. Details of randomised participants: gender, age, family history, speech and language delay, treatment schedule	120
Appendix Table A.2. Primary and secondary outcomes and BC %SS and SRs at each assessment occasion	122
Appendix Table A.3. Parent questionnaire responses	123

LIST OF FIGURES

Figure 3.1. Progression from Stage 1 to Stage 2 visits, adapted from Onslow et al., 2003	42
Figure 4.1. Kaplan-Meier plot of cumulative proportion of subjects who attained Stage 2 by clinic site	59
Figure 4.2. Kaplan-Meier plot of cumulative proportion of 134 subjects who attained Stage 2 by number of clinic visits	60
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.	84
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	86

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.