School of Occupational Therapy, Social Work, and Speech Pathology

Managing fluency and disruptive behaviours in children who stutter: An integrated behavioural and stuttering treatment program

Kerianne Chloe Druker

This thesis is presented for the Degree of
Doctor of Philosophy
of
Curtin University

September 2020

Declaration

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgement has been made. This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

Human Ethics:

The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. The proposed research study received human research ethics approval from the Curtin University Human Research Ethics Committee (EC00262), Approval Number: HRE2017-0652

Signature: …………………………………..

Date: ……..04.09.2020…………………..

Acknowledgments

Friends

Alice Carter, thank you for being my "PhD buddy". I have really enjoyed working with you as a colleague and getting to know you as a friend. I am grateful to you for offering advice and support throughout this journey.

Thank you to all my other special friends for your ongoing encouragement. I am excited to continue celebrating milestones together.

Family

To my husband – Ant, this PhD would not have been possible without your ongoing love and encouragement every step of the way. Thank you for encouraging me to take risks, and reach for the stars. Thank you for travelling the world with me to attend conferences, hopefully this continues for many years into the future! I'm not sure which one of us is more excited for me to finally be called Dr Druker!

To my mom – this PhD is a by-product of all you have invested in me throughout my life. You have worked so hard, and selflessly provided me with the best opportunities - so my achievements are also yours. You are my inspiration.

To my gran – thank you for picking up the phone countless times a day, to allow me to vent and share this journey with you. You are truly the best listener, and I always feel better after hearing your voice and getting your advice.

To my sisters and dad – Cay and Lau, I am so proud of all we have achieved, and I am excited to continue riding through the journey of life together as best friends. Cay, I have to acknowledge your very instrumental role in my pathway to becoming a Speech Pathologist. You have both been extremely supportive throughout this journey. Dad, look forward to making you proud at my graduation!

Abstract

Early developmental stuttering disorders are deemed to be multifactorial in nature, and need to be successfully treated in the preschool years in order to provide children with the best chance of recovery from a potentially life-long disorder. Children who stutter (CWS) as young as three years of age have demonstrated awareness of their disorder, and have been shown to have negative attitudes towards their communication skills. Successful early intervention is required to mitigate the possible negative long term social, academic, occupational and mental health outcomes of living with a chronic stuttering disorder. However, approximately one quarter of CWS do not respond successfully to current early stuttering interventions. In addition, a significant number of children drop out of treatment, highlighting the potential need to support and address other components of the child's developmental and behavioural profile that may hinder or alternately facilitate their engagement with and responsiveness to stuttering treatment.

The behavioural aspect of a child's temperament, self-regulation, has received contemporary theoretical and clinical attention as an important feature in the development and expression of stuttering disorders. However, the impact of the child's self-regulation skills on possibly improving their responsiveness to stuttering treatment has not been determined. Parents act as key agents of change in the management of early stuttering disorders. However, attempts to train parents to support their children's self-regulation skills, as a designated component of stuttering treatment for CWS who present with self-regulation challenges, have not been evaluated.

The present Doctor of Philosophy thesis is written as a series of papers, contributing to understanding the effects of self-regulation difficulties, manifesting in elevated attention-deficit/hyperactivity disorder (ADHD) symptoms, amongst preschool CWS and their parents. The effect on stuttering treatment outcomes and parent and child psychosocial outcomes were explored, and the benefits of addressing self-regulation challenges through clinical intervention were highlighted. The opening paper presented in this thesis comprised a retrospective clinical audit research design, and is the first known study that has quantified the role of ADHD symptoms on stuttering treatment responsiveness. It also determined the proportion of CWS with elevated ADHD symptoms in a large diagnosed clinical population.

Participants were 185 Australian children who had completed stuttering treatment within the last three months prior to the study. The measures included an ADHD measure and relevant clinical variables of interest including age, time between onset of stuttering and therapy commencement, family history of stuttering, sex, pre-treatment stuttered speech severity and typography, number of treatment sessions to successful discharge, as well as post treatment stuttered speech severity. This initial study demonstrated that almost half 49.7% of CWS have concomitant elevated ADHD symptoms. It also determined that elevated ADHD symptoms has a significant and unique predictive effect on stuttering treatment responsiveness. Children with elevated ADHD symptoms were found to require, on average, an additional 25% of clinical intervention sessions to achieve discharge criteria, compared to CWS without these subclinical symptoms.

Research delineating the impact of ADHD symptoms on stuttering treatment responsiveness and the finding that half of CWS in this study met criteria for elevated ADHD symptoms, provided validation for the development of an integrated clinical management plan as the next stage of the research. In addition, the known psychosocial consequences of ADHD symptoms on children and their parents (although subclinical in nature), combined with those documented as a result of stuttering, were deemed important to address in a clinical trial intervention. Therefore, the second paper of this thesis implemented a clinical trial to examine the effectiveness of an integrated evidence-based self-regulation training and early stuttering treatment program on fluency and psychosocial outcomes for the CWS and their parents.

The clinical trial conducted, forming the second paper of the thesis, was quasi-experimental in nature involving 76 CWS and their parents. Thirty-six of these children presented with elevated ADHD symptoms, and were quasi randomised into two groups. The remaining 40 children did not meet criteria for elevated symptoms. All three groups received the standardised stuttering treatment. However, one of the two groups of children with elevated ADHD symptoms, the key intervention group, received an additional evidence-based and parent administered self-regulation component integrated with the stuttering treatment. Stuttering was significantly reduced across all three groups; however, CWS who had elevated ADHD symptoms evidenced a greater reduction in stuttering if they received the integrated program, compared to CWS with elevated ADHD symptoms who received the standardised

stuttering treatment only. At three months post-treatment, stuttering treatment outcomes in the key intervention group were, in fact, equal to CWS without elevated ADHD symptoms. Furthermore, children with elevated ADHD symptoms who received the integrated program group required less stuttering treatment than the comparison group of CWS with accompanying elevated ADHD symptoms. CWS who received the integrated program also demonstrated large and significant reductions in socioemotional difficulty scores and ADHD symptoms, and their parents experienced significant overall improvements in parent practices and family coping strategies. These changes were not observed in the comparison group, and were found to be maintained at follow-up.

In the third paper of this thesis, a qualitative investigation was conducted to assess the social validity, satisfaction and the engagement with the clinical intervention for the families of the children in the integrated treatment condition. In-depth interviews were conducted with eight parents of CWS who received this integrated self-regulation and stuttering program. Questions were open ended and explored parent reflections regarding having a child who stutters with elevated ADHD symptoms, experiences regarding the program, and the perceived social validity of this intervention for these recipients. Interviews were transcribed and analysed thematically. Several major themes were identified including; emotional impact on parents, improvements in child self-regulation, links between stuttering and behaviour, parent self-regulation, impact on family dynamics, and overall positive perceptions of the integrated program. The program was recommended by all participants to any future parents of CWS with concomitant self-regulation issues.

This program of research has identified the impact of childhood self-regulation issues on stuttering treatment outcomes, and demonstrated the effectiveness of an integrated self-regulation and stuttering treatment program for CWS with elevated ADHD symptoms. In addition, qualitative reflections captured from parents provided valuable insights into the clinical benefits of the program. The outcomes of this research have significant implications for future treatments of young CWS and their families.

It is unclear as to whether the impact of self-regulation difficulties and the benefit found when treating these difficulties on stuttering treatment outcomes will translate to other speech and language disorders where there are comorbid and elevated attention and hyperactivity behaviour problems, or whether these findings

will be specific to stuttering, reflecting something fundamental about self-regulatory capacity and stuttering disorders; a possibility that is consistent with a number of current views of stuttering. This remains to be a possible avenue for future research.

List of Publications Arising from this Thesis

Druker, K., Hennessey, N., Mazzucchelli, T., & Beilby, J. (2019). Elevated attention deficit hyperactivity disorder symptoms in children who stutter. *Journal of Fluency Disorders, 59*, 80-90. https://doi:org/10.1016/j.jfludis.2018.11.002.

Druker, K., Mazzucchelli, T., Hennessey, N., & Beilby, J. (2019). Parent perceptions of an integrated stuttering treatment and behavioral self-regulation program. A qualitative thematic analysis. *Journal of Fluency Disorders, 62,* 1-17. https://doi.org/10.1016/j.jfludis.2019.105726.

Druker, K., Hennessey, N., Mazzucchelli, T., & Beilby, J. (2020). An evaluation of an integrated stuttering and parent-administered self-regulation program for early developmental stuttering disorders. *Journal of Speech, Language, and Hearing Research,* 1-19. https://doi.org/10.1044/2020_JSLHR-19-00310.

I warrant that I have obtained, where necessary, permission from the copyright owners to use any third party copyright material reproduced in the thesis (e.g., questionnaires, artwork, unpublished letters), or to use any of my own published work (e.g., journal articles) in which the copyright is held by another party (e.g., publisher, co-author), see Appendix A.

Statement of Author Contribution

The nature and extent of the intellectual input by the candidate and co-authors has been validated by all authors, and can be found in Appendix B.

List of Acronyms Commonly Used in this Thesis

ADHD   Attention deficit hyperactivity disorder

ADHD_IA   Attention deficit hyperactivity disorder, inattentive type

ADHD_HI   Attention deficit hyperactivity disorder, hyperactive-impulsive type

CBQ-EC   Effortful control domain on the Child Behavior Questionnaire

CUSP   Curtin University Stuttering Program

CSTC   Curtin University Stuttering Treatment Clinic

CWS   Children who stutter

CWNS   Children who do not stutter

eADHD   Elevated ADHD symptoms

EBPS   Evidence based parenting support

PAFAS   Parenting and Family Adjustment Scales

SDQ   Strengths and Difficulties Questionnaire

%SS   Percentage of syllables stuttered

Table of Contents

Chapter 1

Overview of Doctor of Philosophy

Chapter 1: Overview of Doctor of Philosophy

This thesis contains novel findings that have the potential to impact delivery of intervention for preschool stuttering disorders. It commences by providing a summary of the evidence for the role of self-regulation in early stuttering, supported by contemporary theoretical models of early stuttering. The overall poorer self-regulation skills in children who stutter (CWS) compared to their fluent peers is described. In addition, preliminary research indicating that self-regulation difficulties may impact negatively on stuttering treatment outcomes is explored. The thesis is presented as a series of three individual papers, culminating in a discussion of the project's strengths and limitations, and directions for future research. Throughout the thesis, studies include CWS of preschool age, given the recognised contemporary importance of providing efficient and successful intervention as close to the onset of stuttering as possible.

Study 1 is the second largest, known retrospective clinical audit of preschool CWS who have received stuttering treatment. The paper identifies the proportion of preschool CWS who present to stuttering clinics with self-regulation challenges, operationalised as elevated symptoms of attention deficit hyperactivity disorder. This is the second time the proportion of CWS with concomitant self-regulation challenges has been quantified in the literature, and the sample size in the present study is greater than that of the previous reported study (Donaher & Richels 2012). The paper is also the first to identify an aspect of temperament that significantly predicts stuttering treatment outcomes, highlighting the need to consider this in treatment planning. Findings, therefore, provide support for a prospective clinical trial to focus on self-regulation challenges in addition to stuttering.

The retention of clients in the clinical trial was strong with an attrition rate of 8.1%. This compares favourably with previous case file audits of preschool stuttering treatment, where attrition rates ranged from 15% to 57% (Kingston et al., 2003; McCulloch et al., 2017). This suggests that the nature of the stuttering intervention delivered in the current study is acceptable, positive and feasible for the families involved in this body of research (Kingston et al., 2003; McCulloch et al., 2017).

A large scale three-armed quasi-experimental prospective clinical trial was employed in the second study of this thesis. Similar to Study 1, a comparable proportion of preschool CWS with concomitant self-regulation issues was obtained,

giving confidence to the notion that a substantial number of CWS present with these difficulties. In addition, the impact of self-regulation issues on stuttering treatment outcomes were found to be similar across the two studies. A novel integrated self-regulation and stuttering intervention was implemented in the target intervention group, and compared to two control groups (based on child profile as well as intervention type administered). The self-regulation component comprised the Triple P Positive Parenting Program (Sanders, 2012), adapted for the developmental stuttering population families. The stuttering component of the treatment comprised direct and indirect stuttering treatment principles, as implemented at the Curtin University Stuttering Treatment Clinic. Positive psychosocial outcomes and enhanced fluency outcomes were demonstrated in the key intervention group, with large effect sizes. Such psychosocial and fluency benefits were found to be robust and durable over the three-month follow up period. A similarly low attrition rate to that reported in Study 1 was obtained.

Given the novel nature of the integrated self-regulation and stuttering intervention, it was important to obtain the participants' points of view, in order to gain their reflections and insights surrounding their experiences as caregivers implementing this intervention with their children. Therefore, in Study 3, qualitative data was collected to augment quantitative outcomes yielded in the intervention study, strengthening the acceptability and social validity of the novel intervention (Palinkas et al., 2011). The concurrent benefits of addressing self-regulation and stuttering in naturalistic daily parent-child interactions were highlighted by the parents in this study. The qualitative themes that emerged focused mainly on parent and child self-regulation, and highlighted the link between stuttering and self-regulation in daily interactions. The clinical benefits of addressing the stuttering disorder holistically, and treating the child's stuttering disorder in the context of the entire family system was elucidated. There were improved positive interactions between the child and other family members reported. This thesis employed mixed quantitative experimental and qualitative methodologies in the studies and demonstrated complementary findings across both research methodologies.

The positive impact of the integrated intervention on both fluency and psychosocial outcomes, assessed by strongly validated measures of child and parental mental health, underscores the multifaceted and complex nature of developmental stuttering disorders.

Chapter 2

Introduction

Chapter 2: Introduction

Stuttering is a multifactorial communication disorder that involves fluctuating and involuntary motoric breakdowns in fluent speech production, resulting in interruptions to the rhythm and flow of fluent speech (Manning & DiLollo, 2018). This complex communication disorder may impact an individual in many areas of life, far beyond the observable and physical speech disfluencies (Beilby, 2014).

Models of Developmental Stuttering

Throughout history, theories and models have attempted to elucidate factors that contribute to the onset of stuttering, its manifestation, and possible factors hindering or promoting recovery. Many early accounts of the emergence of stuttering lacked an empirical, scientific base and were primarily based on anecdotal accounts (Bloodstein & Bernstein Ratner, 2008). These included: abnormalities of the speech apparatus, psychogenic causes, and negative parental reactions to normal childhood disfluencies (e.g., Plankers, 1999). In contemporary literature, however, models have shifted to focus more on the neurobiological, multidimensional nature of developmental stuttering disorders.

Multifactorial models, widely accepted in contemporary clinical practice, are described below in order to provide a contextual background for the integrated stuttering and behavioural self-regulation intervention that was implemented and evaluated in this body of research. The models selected recognise and capture the role of emotional factors, including self-regulation, in the manifestation of stuttering symptoms. Self-regulation encompasses behavioural and/or emotional regulation, and refers to the predominantly volitional cognitive and behavioural responses important for the completion of goal-directed activities (Blair & Diamond, 2008; Posner & Rothbart, 2009). The models chosen include: the dual diathesis-stressor model (Choi et al., 2016; Conture & Walden, 2012; Walden et al., 2012), the multifactorial dynamic pathways theory (Smith & Weber, 2017) and the demands and capacities model (DCM; Starkweather, 1987).

Dual Diathesis-Stressor Model

In developing the dual diathesis-stressor model, Walden et al. (2012) recognised the need to identify factors that initiate, exacerbate and perpetuate stuttering disorders, in light of the extensive negative psychosocial consequences of chronic stuttering (Beilby, 2014). This model proposes that external or

environmental emotional and speech-language stressors to which the individual is exposed, interact with emotion and speech-language diatheses (individual differences), leading to disruptions in speech fluency. Emotional stressors are different features of a situation that elicit emotion (e.g., sibling rivalry over a toy), and speech-language stressors are requirements for effective communication that vary in different situations (e.g., complexity of linguistic task). This model is of primary interest for the current body of research given its focus on the role of emotional factors in the manifestation of stuttering in the individual.

The emotional components of the dual diathesis-stressor model comprise two aspects: emotion reactivity and emotion regulation (Walden et al., 2012). Reactivity refers to emotional responsiveness to environmental stimuli that varies between individuals in terms of latency, intensity and duration (Rothbart et al., 2001). For example, a child with higher reactivity may experience more emotional outbursts, become more easily excited, become more upset if something unexpected occurs, as well as have difficulty calming down when upset. A child with lower reactivity may be perceived as easy-going and be less likely to experience such emotional outbursts (Rothbart et al., 2001).

Emotion-regulation refers to implicit and/or effortful processes that work to modulate emotional reactivity, including behavioural approach, inhibition, withdrawal and executive or effortful attention (Rothbart et al., 2001; Sheppes et al., 2015). A child with weaker emotion-regulation skills may be more impulsive, and have greater difficulty regulating attention and managing emotions. A child with stronger emotion regulation skills may be better able to regulate attention, persist with tasks, and be less impulsive.

According to this model, emotional reactivity, emotional regulation, and their combined effects can impact the severity and frequency of stuttering of a preschool CWS (Walden et al., 2012). Specifically, a predisposition for increased emotional reactivity and lower emotional regulation is anticipated to be associated with increased stuttering (Walden et al., 2012). For example, a young child with increased reactivity and weaker emotional regulation skills may display increased tantrums and emotional outbursts in response to disciplinary boundaries set by parents. This proclivity for greater emotional outbursts is likely to increase severity and/or frequency of stuttered speech in these moments of emotional distress. This proposed causal mechanism is based on the authors' diathesis-stress approach to stuttering,

whereby emotional and language stressors are proposed to exacerbate individual emotion and language diatheses, leading to disruptions in fluency.

To test the dual diathesis-stressor model, Walden et al. (2012) observed behaviours in CWS following exposure to emotionally evoking experimental conditions, including "overheard" happy, neutral and frustrating conversations. CWS were then asked to retell a narrative using picture book illustrations. Emotional reactivity and regulation behaviours were coded during the overheard conversations and the narratives. Emotional reactivity was coded based on expression of positive (e.g., smiles) or negative emotions (e.g., frowns). Emotional regulation was coded based on demonstration of regulatory behaviours (e.g., self-stimulation with or without visual attention and social looks at the examiner).

It was found that CWS who exhibited increased negative emotion without concomitant self-regulatory attempts displayed more stuttering. In contrast, CWS who exhibited both increased negative emotion, and increased self-regulatory attempts, stuttered less. Furthermore, stuttering increased in CWS following exposure to both emotionally evoking experimental conditions, providing empirical support for this model that posits unchecked emotional arousal (whether positive or negative) impacts fluency (Walden et al., 2012). The diathesis-stress approach therefore indicates that at least some component of stuttering management would benefit from a focus on managing environmental stressors, and also potentially, the emotional responses to those stressors on the part of the child.

Multifactorial Dynamic Pathways Theory

The multifactorial dynamic pathways theory (Smith & Weber, 2017) was an advancement of a model developed by Smith (1999), which was one of the first models to suggest that dynamic rather than more static difficulties confined to the speech motor control system of individuals who stutter, contributed to stuttering events. The advanced multifactorial dynamic pathways theory focuses on the multiple events occurring in the window of time when stuttering emerges and recovery or persistence ensues. Emphasis is placed on the diversity and interplay of genetics (Drayna & Frigerios-Domingues, 2017; Fagnani et al., 2011), epigenetics (Drayna & Frigerios-Domingues, 2017; Fagnani et al., 2011), motoric factors (MacPherson & Smith, 2013; Walsh et al., 2015), linguistic factors (Spencer & Weber-Fox, 2014), as well as emotional and psychosocial factors (Jones et al., 2014b) in determining the prognosis of early stuttering disorders for individuals. The

ultimate goal of this model is to develop an "understanding of how best to aid children in finding the pathway to recovery" (Smith & Weber, 2017, p. 2485).

The current PhD research project focuses on the potential role of emotional demands represented in this model. These demands are considered to be one of the key factors that potentially constrain the complex speech motor speech system and extend it beyond the capacity for fluent speech production, impacting stuttering onset, manifestation and prognosis (Smith & Weber, 2017). This model is similar to the dual-diathesis stressor framework (Walden et al., 2012) in that it recognises emotional and linguistic factors as key influences in the developmental stuttering disorder trajectory.

Demands and Capacities Model

The DCM proposed by Starkweather in 1987, was aimed at addressing the development of stuttering in young children. Starkweather (1987) positioned the DCM to advance the theory of developmental stuttering, facilitate experimental research, and provide support for clinical assessment and intervention (Starkweather & Ridener Gottwald, 1990). Treatment based on the DCM has been demonstrated to be successful for early developmental stuttering disorders (De Sonneville-Koedoot et al., 2015). The DCM model proposes that CWS have a genetic predisposition or diathesis for disfluent speech production within their speech motor system, in line with the multifactorial dynamic pathways theory. Stuttering begins and is then precipitated by an imbalance between the demands of the external environment and the child's internal capacities for fluent speech. This impacts the speech motor planning and execution abilities necessary for fluent speech production (Starkweather, 1987). Demands in this model refer to external or environmental dimensions/pressures including: linguistic (e.g., use of semantically and syntactically complex utterances), motor (e.g., speaking when listeners are in a hurry), cognitive (e.g., level of metalinguistic skills required in a task), and socio-emotional factors (e.g., child possessing low self-regulation ability and high levels of emotional volatility).

The DCM model aligns with recommendations made by authors of the multifactorial dynamic pathways theory, in that this model supports a focus on strengthening the child's capacity for fluency by addressing certain demands that may challenge their capability for fluent speech production (Starkweather, 1987). An analogy by Starkweather and Ridener Gottwald (1990) demonstrated the importance

of addressing such demands to support successful early intervention in young CWS. These authors compared fluency management without consideration of environmental demands to a "commuter running after the departing train, [who] fights a losing battle unless the train can be slowed" (p. 145).

In summary, the different multifactorial models have acknowledged the relevance of emotion and environmental factors as potential influences in early developmental stuttering. These models point to the possible importance of integrating these components into early stuttering management, with the potential to address these factors in order to facilitate or enhance recovery from developmental stuttering.

Intervention Approaches for Developmental Stuttering

Contemporary models of early developmental stuttering have highlighted the multifaceted nature of early developmental stuttering, and approaches to early stuttering treatment can be evaluated in this perspective. Previous published reviews of preschool stuttering treatments have broadly divided treatment approaches into direct and indirect categories (Donaghy & Smith, 2016).

Direct treatment approaches target the speech motor system directly and require CWS to make specific changes in their speech (Onslow et al., 2003). Direct approaches often include operant methods, whereby verbal contingencies are provided following fluent and stuttered speech (Donaghy & Smith, 2016; Onslow & O'Brian, 2013). Such treatment programs, for example, the Lidcombe Program, aim to promote fluency at a single word level with gradual increases in utterance length, while maintaining fluent speech production (Onslow et al., 2003). Theoretically, reductions in stuttering using direct treatment approaches occur due to operant methods, as well as motoric practice of stutter-free speech, or a combination of these two factors (Onslow & O'Brian, 2013). Less focus appears to be given to the multifactorial nature of the disorder, recognised in contemporary theoretical models. Treatment goals in these approaches are predominantly guided by fluency progress and reduction in stuttered speech severity (Onslow & O'Brian, 2013; Onslow et al., 2003).

Multifactorial treatment approaches, which have been referred to as indirect approaches (Donaghy & Smith, 2016), predominantly work to address or reduce demands set by the environment to promote the child's capacity for fluent speech

production, with the aim of reducing stuttering (Donaghy & Smith, 2016)[1]. The theoretical basis for these treatment approaches is primarily based on the DCM (Donaghy & Smith, 2016). In these approaches, e.g., RESTART-DCM (Franken & Putker-de Bruijn, 2007) and Palin Parent Child Interaction Treatment (Millard et al., 2008; Millard et al., 2018), treatment may focus on altering parent behaviours, interactions and family routines that may increase environmental demands. For example, parents may be instructed to modify their interaction style to facilitate fluency by demonstrating supportive listening, follow the child's lead, slow their speech rate, and reduce complexity of language during conversation interactions with the CWS (Millard et al., 2008).

The common factors approach is an overarching set of principles used in research of health and psychological interventions, which is relevant when exploring commonalities between preschool stuttering treatment approaches (Wampold, 2015). In essence, this approach posits that commonalities across treatments, as opposed to unique ingredients, are responsible for treatment success (Benish et al., 2011; Wampold, 2015; Webb et al., 2010). There are multiple common factors of direct and indirect/multifactorial treatment approaches that warrant highlighting (Yaruss, 2019). These include, involvement of parents in all aspects of the intervention process, parent education, encouraging parents to support their children to successfully convey their communicative messages, parent training to manage their children's stuttering beyond the clinic environment in a range of daily situations, goals of stuttering reduction and associated improved fluency, as well as parent acknowledgment of stuttering in a neutral way (Yaruss, 2019).

The nature of the common elements encompassed within different early stuttering intervention approaches implies that the notion of direct and indirect/multifactorial treatment in future clinical practice may not warrant the contrastive viewpoints; rather, individualising and adapting key aspects of successful treatments to each child and family in a flexible way may be beneficial and

---

[1] The direct versus indirect treatment distinction can have a different meaning for some authors, based on whether the treatment is with the child themselves (e.g., targeting their speech motor system or any other aspect of the child's behaviour) versus a treatment approach that works through the parent or caregiver rather than the child (and therefore only indirectly targets the child). In the present thesis, a direct approach refers to any treatment that targets specifically the child's speech production system (e.g., using operant methods), whereas indirect treatment seeks to address the child's fluency through targeting other factors (e.g., environmental demands) than the child's stuttering behaviour, whether that is primarily through the parent or involves the child as well.

worthwhile (Yaruss, 2019). This more flexible and individualised approach to stuttering treatment aligns with multifactorial models of stuttering, as well as current trends in individualised medicine. It may also allow for further consideration of additional environmental demands and/or personal capacities, as part of holistic treatment for development stuttering disorders.

Evidence for Preschool Stuttering Treatment Approaches

Historically, reviews of stuttering treatment have evaluated the evidence base and recognised direct approaches to stuttering treatment to be most effective in reducing the severity of stuttered speech for preschool CWS (Blomgren, 2013; Nye et al., 2013; Sidavi & Fabus, 2010). However, there has been limited research to date that has directly compared the effectiveness of direct and multifactorial/indirect treatment approaches. The comparative effectiveness of these two treatment approaches in reducing stuttered speech severity for preschool age CWS was first demonstrated by a recent randomized controlled trial of preschool stuttering intervention (De Sonneville-Koedoot et al., 2015). In this clinical trial, findings demonstrated that there were no statistically significant differences in the percentage of children who had recovered from stuttering at 18 months follow up (i.e., $\leq$ 1.5%SS) following direct treatment (65/85; 76.5%) and indirect treatment (65/91; 71.4%). Furthermore, stuttered speech severity was also equal in both treatment groups at 18 months post-treatment, suggesting equal effectiveness of the treatment approaches. It is worth noting that despite the demonstrated equal effectiveness of direct and indirect/multifactorial stuttering interventions, around 25% of CWS did not have their stuttering disorders remediated at follow-up, regardless of the treatment approach implemented.

The preliminary effectiveness of more flexible preschool stuttering treatment approaches, whereby direct and indirect treatment principles are flexibly combined, has been demonstrated in relatively small scale studies to date (Druker et al., 2019; Yaruss et al., 2006), and requires further replication.

The comparative effectiveness of direct and indirect treatment approaches has been acknowledged in the most recent review of preschool stuttering treatment (Donaghy & Smith, 2016). A notable final recommendation made in this review suggested that future research regarding the contributing components of early stuttering treatments may be instrumental in promoting clinical problem solving for CWS who may not respond successfully to previous stuttering treatments, or who

have concomitant disorders that may influence the manifestations of their stuttering disorders.

Importance of Early Stuttering Intervention

Contemporary stuttering treatment approaches outlined above have been found to be effective in reducing stuttered speech severity for a large proportion of children, but approximately one quarter of preschool-age CWS appear not to have their disorder successfully remediated following treatment, whether the treatment implemented is direct or indirect/multifactorial (De Sonneville-Koedoot et al., 2015). Neuroimaging data indicates differences between CWS and children who do not stutter (CWNS) in the auditory and motor areas of the brain, as well as in the cortical and subcortical circuits of the brain, including the supplementary motor area and putamen, at even the earliest stages of stuttering. Chang (2014) concluded that the brain regions found to differ in CWS are primarily those that experience active change and growth during early childhood. Chang (2014), therefore, suggested that young CWS are more likely to respond positively to early intervention, given the plasticity of affected brain areas during early development. This highlights that providing successful early intervention is important in the early time window after stuttering begins, before the chance of recovery reduces. In this light, investigation into how to maximise early stuttering treatment outcomes for children who do not respond to available stuttering treatments is desirable.

Psychosocial Consequences of Living with Stuttering.

There are notable possible negative psychosocial consequences resulting from living with a stuttering disorder (Beilby, 2014). Families of CWS have reported family conflict, increased emotional burden, and difficulties managing the frustrations experienced by their children due to their stuttered speech difficulties (Erickson & Block, 2013). Children as young as three years-of-age have been shown to have awareness of their disfluency, and may develop a negative attitude towards their speech by six years of age (Vanryckeghem et al., 2005). In addition, research examining the impact of early stuttering on peer interactions has found that preschoolers who stutter are more likely to have difficulty with conflict resolution, and are less likely to lead or participate in pretend play (Langevin et al., 2009). These findings highlight the potential negative impact of stuttering on early pragmatic and peer relationship development.

The long-term social and emotional outcomes for CWS at ages three-, five- and eleven-years indicated that CWS at age three presented with significantly higher social and emotional problems than their fluent peers, including difficulties with behavioural, emotional and social development (McAllister, 2016). By age five and age eleven, CWS were more likely than their fluent counterparts to have clinical levels of conduct problems, emotional problems, peer problems and hyper-activity (McAllister, 2016). It is also recognised that persistent stuttering beyond childhood may have adverse outcomes on educational attainment (O'Brian et al., 2011), vocational choice (Klein & Hood, 2004), romantic relationships (Van Borsel et al., 2011), personal self-esteem (Klompas & Ross, 2004), and overall quality of life (Carter et al., 2017).

These extensive negative psychosocial outcomes demonstrate the significance of prioritising treatment efficacy in the early stages of stuttering. This is particularly important considering the proportion of CWS who have not had their disorders remediated following treatment (De Sonneville-Koedoot et al., 2015), the low recovery rates post 12 months onset (Carey et al., 2020b; Reilly et al., 2013), and the documented high attrition rates from treatments (e.g., McCulloch et al., 2017; Trajkovski et al., 2019). In light of this, researchers have begun to discuss the importance of acknowledging and treating multifactorial factors within a child's profile that may hinder or support recovery from stuttering, particularly for children who may be resistant to available stuttering treatment approaches (Donaghy & Smith, 2016; Smith & Weber, 2017). For example, emotional factors, including self-regulatory skills, have been flagged as a potential factor to facilitate improved fluency in CWS (Smith & Weber, 2017).

Self-Regulation and Developmental Stuttering Disorders

The possible impact of self-regulatory skills on developmental stuttering onset, manifestation and/or prognosis has been discussed in research and clinical fields (Ambrose et al., 2015; Kraft et al., 2014; Zengin-Bolatkale et al., 2018b). Self-regulation is important in childhood stuttering because the child is facilitated to inhibit the stuttered speech behaviour in favour of a more desirable fluent speech behaviour, as well as attend to treatment strategies within and beyond the clinical environment. Children with heightened emotional reactivity are likely to respond to environmental changes with increased emotional volatility. Children with self-regulation challenges have difficulty tempering these emotional responses (Posner &

Rothbart, 2009). They, therefore, may display behaviours such as intense emotional arousal in response to negative and/or positive environmental stimuli, impulsive behaviour and difficulty shifting and focusing attention (Miller et al., 2004; Posner & Rothbart, 2009).

The DCM can be used to support the role of self-regulation in fluent speech production, as well as to demonstrate the possible relevance of considering self-regulation as a component of stuttering intervention. When children respond with increased impulsivity and volatility to environmental stimuli, or have difficulty shifting and focusing attention appropriately, there is a greater discrepancy between capacities and demands (Starkweather, 2002; Starkweather et al., 1997). For example, a child who is overly restless may experience a drain in motoric resources, and an overly emotional and reactive child may experience excessive depletion of emotional resources (Starkweather et al., 1997). This depletes resources available for fluent speech production, as well as for attending to stuttering treatment strategies within and beyond the clinic. If the child's capacity to react to environmental stimuli with less impulsivity and emotional volatility is enhanced, resource demands for fluency may be freed up, preventing the motoric practice of disfluent speech production (Onslow & O'Brian, 2013). This points to the possible importance of addressing self-regulation difficulties as a useful component of stuttering intervention, at least for CWS challenged with some level of self-regulation difficulties.

A large number of studies that have examined self-regulation in preschool CWS have been observational in nature, and have compared self-regulation characteristics between CWS and CWNS. It has been demonstrated that preschool CWS, as a group, present with poorer self-regulation skills and increased emotional reactivity compared to their fluent peers. These broad group differences were mostly consistent across a range of research methodologies employed, including parental report (e.g., Anderson et al., 2003; Eggers et al., 2010; Embrechts et al., 2000; Howell et al., 2004; Karrass et al., 2006), behavioural coding measures (e.g., Arnold et al., 2011; Jones et al., 2014c; Ntourou et al., 2013; Schwenk et al., 2007; Walden et al., 2012), and physiological testing of the autonomic nervous system (Arnold et al., 2011; Jones et al., 2014a; Zengin-Bolatkale et al., 2015). Beyond the overall group differences that have been demonstrated in research to date, it has also been concluded that poorer emotional regulation attempts and increased emotional

reactivity are associated with more frequent stuttering in CWS (e.g., Arnold et al., 2011; Walden et al., 2012). These research findings provide support for potential consideration of emotional regulation skills of CWS as part of a holistic stuttering management program, with possible benefits to fluency improvements.

Temperament and Stuttered Speech Severity

Extending beyond research whereby differences in self-regulation skills between CWS and CWNS have been examined, a hallmark study investigated factors that may be related to stuttering severity (Kraft et al., 2014). Factors included sex, age, socio-economic status, home environment, negative life events and temperament (Kraft et al., 2014). Of all factors tested, the only factor found to be significantly related to stuttering severity was the temperament construct of effortful control on the Child Behavior Questionnaire (CBQ; Rothbart et al., 2001). Effortful control is an index of self-regulation, that refers to the ability to inhibit a dominant response in favour of activating a subdominant/more desirable response (Rothbart et al., 2003). A significant negative relationship was found between stuttered speech severity ratings and effortful control, demonstrating a statistically significant association between the index of self-regulation on the CBQ and stuttered speech severity. Similar findings were yielded by a replication study with a larger sample size (Kraft et al., 2019), and provide further support for the consideration of self-regulation skills in stuttering management, to potentially enhance reductions in stuttered speech severity for CWS.

Attention Deficit Hyperactivity Disorder Symptoms in CWS

Self-regulation issues experienced by CWS may be similar to those exhibited in children with a diagnosed attention-deficit/hyperactivity disorder (ADHD). A core deficit of ADHD is self-dysregulation, characterised by attention and/or inhibitory control difficulties (Barkley, 2013). The majority of studies that have examined group differences in temperament characteristics between preschool CWS and CWNS have found that CWS do indeed display specific challenges with attentional allocation (e.g., Eggers et al., 2012) and inhibitory control (e.g., Anderson & Wagovich, 2010; Eggers et al., 2013). However, contradictory findings have been found where differences in these temperament characteristics were not observed (e.g., Kefalianos et al., 2014). Limitations of parent report methodology have been noted, possibly contributing to contradictory findings presented. It is possible that parents of CWS may perceive greater difficulties in temperament features of their

child due to the distress and discomfort this disorder is known to invoke in parents (Langevin et al., 2010; Plexico & Burrus, 2012). Furthermore, the specific parent report measures of child temperament are not consistent across all studies.

Researchers have also examined the potential overlap in behavioural characteristics between CWS and ADHD, and found a greater proportion of CWS to meet the clinical threshold for ADHD symptoms, compared to CWNS (Alm & Risberg, 2007; Felsenfeld et al., 2010). Furthermore, the proportion of CWS with elevated ADHD symptoms, although subclinical in nature, has been found to be 58% ($n = 36$; Donaher & Richels 2012). Replication of this finding, with a larger sample size, is warranted given the potentially large proportion of CWS who present to clinic with these co-occurring behavioural challenges.

Inattention and inhibitory control difficulties, reflective of ADHD-like symptoms, have been suggested to influence stuttering manifestation. Eggers et al. (2013) suggested that lower inhibitory control may "increase the amount of emotional arousal some [CWS] experience in stressful situations, impacting their stuttering symptoms" (Eggers et al., 2013, p. 8). One study to date (Riley & Riley, 2000) has examined the impact of attending difficulties on stuttering treatment responsiveness. These authors found that CWS with comorbid "attending disorders" at pre-treatment that were not addressed, had a significantly greater chance of having an "unacceptable level of stuttering" (p. 191) 2- to 4-years after treatment termination, compared to children without an "attending disorder". Further evaluation of adjunctive or integrated management of ADHD-like symptoms, as a component of preschool stuttering treatment, may shed light on the potential importance of addressing these symptoms to strengthen stuttering treatment responsiveness and outcomes for this possible subgroup of CWS.

Psychosocial Impact of Self-Regulation Challenges

A significant proportion of preschool CWS have been found to present with poor self-regulation skills (Donaher & Richels, 2012), known to manifest in elevated ADHD symptoms (Barkley, 2013; Huguet et al., 2019; Reid et al., 2005; Spencer et al., 2011). Poor self-regulation skills place these children at a possible increased risk of negative long term outcomes in a range of life areas, including education, socialisation, employment, and mental health (Moffitt et al., 2011; Tsukayama et al., 2010). Therefore, if these symptoms are not addressed as part of the holistic management of preschool stuttering disorders, they may further compound the

already negative psychosocial outcomes of developmental stuttering for CWS and their families (Beilby, 2014; Langevin et al., 2010; Millard & Davis, 2016; Vanryckeghem et al., 2005).

Parent Role in Early Intervention

It is well documented that parents play a crucial role in the management of developmental stuttering disorders (Franken & Putker-de Bruijn, 2007; Millard et al., 2008; Onslow et al., 2003; Yaruss et al., 2006). Unfortunately, parents of CWS have reported a myriad of negative emotional consequences resulting from their child having a stuttering disorder, including feelings of guilt, anxiety, and uncertainty about how to manage their children's disorder (Langevin et al., 2010). Therefore, provision of holistic support to parents in managing their children's potentially variable stuttering disorder is important. In doing so, the clinical responsiveness and successful outcomes of early developmental stuttering treatments may also be facilitated.

Management of ADHD-Like Symptoms through Parenting Support

The potential challenges posed by a child presenting with elevated ADHD symptoms, as well as a clinically diagnosed stutter, suggests the possible need for an integrated clinical management program. In theoretical models of self-regulation (Blair & Ursache, 2011; Sameroff, 2010) parents have been acknowledged as key agents of change in development of self-regulation skills in their children. These models provide theoretical support for training parents to support the development of self-regulation skills in CWS, particularly those that may be identified to have weaker self-regulation skills. Training parents to support the development of self-regulation in their children, using evidence-based parenting support (EBPS), has been shown to reduce behaviours resembling ADHD, including inattentive or impulsive behaviours (Mazzucchelli, 2018; Sanders & Mazzucchelli, 2013). As such, although poor self-regulation skills may place a child at an increased risk of negative psychosocial outcomes (Moffitt et al., 2011), training parents to improve self-regulation in CWS can moderate the impacts of self-regulation difficulties (Deater-Deckard, 2014; Sameroff, 2010; Ullsperger et al., 2016).

Evidence Based Parenting Support for Improving Child Behaviour

EBPS programs founded on social learning theory and behavioural science (Sanders & Mazzucchelli, 2018) aim to provide support to parents in the development of self-regulation skills in their children, through the promotion of

consistent and positive parenting skills. Comprehensive reviews of the evidence in the area of EBPS (e.g., Gray et al., 2018; Pidano & Allen, 2015; Sanders et al., 2014) have indicated that EBPS can be effective in improving both child behaviour and parenting outcomes. Positive psychosocial outcomes for children include promotion of self-regulation skills; involving reduction in disruptive child behaviours and improved social and emotional development (Gray et al., 2018; Pidano & Allen, 2015; Sanders et al., 2014). Positive parental outcomes include the development of adaptive parenting practices, reduction of ineffective discipline techniques, and improved maternal mental health (Gray et al., 2018; Pidano & Allen, 2015; Sanders et al., 2014). Adaptions of EBPS to particular populations have been successful in yielding similar positive parent and child psychosocial outcomes, through development of resources that flexibly adapt the EBPS principles to the specific disorder of interest (e.g., Hoath & Sanders, 2002). As such, it is hypothesized that EBPS principles may be effective for parents of preschool CWS, to support them to improve self-regulation skills in their CWS with comorbid difficulties in this area.

Rationale for Current Research

Self-regulation skills have been investigated as a potentially important factor in the course of developmental stuttering disorders. Specifically, CWS have been found to display greater difficulties in self-regulation skills than their fluent peers (including greater difficulties with attentional regulation and inhibitory control). In addition, significant relationships between self-regulation and stuttering frequency have been established. Finally, the possible impacts of self-regulation difficulties on the manifestation of developmental stuttering disorders have been proposed.

Proportion and Effect of Self-Regulation Challenges on Stuttering Treatment. One previous study has investigated the proportion of CWS with self-regulatory challenges manifesting in elevated ADHD symptoms (Donaher & Richels, 2012). The large proportion of CWS found to present with comorbid ADHD symptoms requires replication with a larger sample size. Research has also provided preliminary evidence that such symptoms may compromise stuttering treatment outcomes (Riley & Riley, 2000). This highlights the need to quantify the potential impact of ADHD symptoms on stuttering treatment responsiveness and outcomes.

Addressing Self-Regulation in Early Stuttering Intervention. Theoretical support exists for addressing self-regulation skills in a subgroup of CWS who are challenged with self-regulation difficulties, given the potential influence of these

skills on stuttering manifestation (Walden et al., 2012). Furthermore, the presence of self-regulation difficulties may impact a child by challenging their ability to appropriately regulate their emotions and behaviours on a day-to-day basis (Moffitt et al., 2011). This may compound the potentially adverse long-term psychosocial impacts of a chronic stuttering disorder. As such, addressing these symptoms as part of a holistic and multifactorial intervention may promote positive psychosocial development for these children and their families, in conjunction with projected enhancement and maintenance of fluency improvements over time.

The importance of supporting child self-regulation in developmental stuttering has been recognised in other multifactorial stuttering treatment approaches. For example, the RESTART-DCM (Franken & Putker-de Bruijn, 2007) approach has acknowledged the importance of considering the child's temperament in early stuttering management. Strategies to address possible excessive emotional demands in reactive children have included: the promotion of safety and predictability within the child's home environment, problem solving and adaptive conflict resolution strategies, as well as labelling of emotions (Franken & Putker-de Bruijn, 2007). Parental problem solving has also been recommended to enable parents to better cope with a reactive child and their self-regulation capacity (Franken & Putker-de Bruijn, 2007). Within the Palin Parent Child Interaction treatment, strategies have been provided to parents to support them to regulate their children's emotions, with focus on improving routine and predictability (Millard et al., 2008).

The possible impact of improving child self-regulation skills on stuttering treatment outcomes has been examined in a preliminary study underpinning the current PhD research project, whereby an integrated self-regulation and stuttering treatment program was piloted (Druker et al., 2019). Study findings were positive in terms of significantly improved psychosocial outcomes for both parents and children, including reduction in child socioemotional problems, as well as improved parenting practices and family functioning as a whole. However, self-regulation principles did not appear to have a statistically significant impact on stuttering treatment outcomes in this study. In addition, the self-regulation component was not evidence based, and CWS who received the integrated program did not necessarily meet criteria for having comorbid self-regulation challenges.

The importance of considering emotional factors in developmental stuttering has been recognised by some multifactorial treatment approaches and in a

preliminary pilot study. However, the effectiveness of implementing EBPS to address emotional factors as a component of early stuttering management, with the aim of maximising stuttering treatment outcomes for CWS particularly challenged with comorbid self-regulation difficulties, requires further investigation.

Objectives

The overarching aim of the current research project was to determine if strengthening self-regulation skills would result in improved stuttering treatment outcomes, for CWS specifically presenting with comorbid self-regulation challenges. To address this aim, three studies were employed with the following key objectives.

Study 1 aimed to: (a) identify the proportion of CWS who have elevated ADHD symptoms, (b) determine if elevated ADHD symptoms predicted responsiveness to stuttering treatment, (c) investigate potential differences between CWS with and without elevated ADHD symptoms, in terms of pre-treatment stuttering features, demographic variables, and treatment outcome data.

The primary aim of Study 2 was to (a) examine the impact of an integrated self-regulation and stuttering treatment program on stuttering treatment outcomes, for a subgroup of CWS who met criteria for comorbid self-regulation challenges. Secondary aims in this study were to (b) examine the impact of the integrated program on child and parent psychosocial outcomes, and (b) compare presentation characteristics between groups of CWS with and without self-regulation challenges/elevated ADHD symptoms. The Curtin University Stuttering Program (CUSP) was the stuttering treatment implemented in this controlled clinical trial (Druker et al., 2019). This treatment approach is based on a combination of direct and multifactorial/indirect stuttering treatment principles. Therefore, it was hoped that the findings from this body of research may add to the evidence base regarding the preliminary effectiveness of a preschool stuttering treatment that flexibly combines principles of direct and indirect/multifactorial treatment approaches. As such, the final secondary aim of this study was to (c) determine the effectiveness of the stuttering treatment implemented in this clinical trial.

Study 3 employed qualitative research methodology to gain a deeper understanding of the experiences of parents of children who received the integrated self-regulation and stuttering program, to gather insights into the acceptability and social validity of the novel intervention within this population.
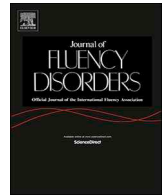
Chapter 3

Paper 1: Elevated attention deficit hyperactivity disorder symptoms in children who stutter

Elevated attention deficit hyperactivity disorder symptoms in children who stutter

Kerianne Druker[a], Neville Hennessey[a], Trevor Mazzucchelli[b], Janet Beilby[a,*]

[a] School of Occupational Therapy, Social Work and Speech Pathology, Curtin University, Western Australia, Australia
[b] School of Psychology, Curtin University, Western Australia, Australia

A B S T R A C T

Purpose: This study described the proportion of children who stutter who exhibit Attention Deficit Hyperactivity Disorder (ADHD) symptoms, manifesting in inattentive and hyperactive/impulsive behaviours. Children who stutter with these challenging behaviours may not respond as quickly and successfully to stuttering treatment. A preliminary exploration of differences in treatment responsiveness for children with and without ADHD symptoms was undertaken.
Method: Participants were 185 preschool children who stutter who had completed stuttering therapy within 3 months prior to study commencement. Differences between groups of children who stutter with and without elevated ADHD symptoms were investigated, in terms of pre-treatment stuttering features (stuttering severity and typography), demographic variables (age at onset, time between onset and commencement of therapy, family history and sex) and treatment data (post-treatment stuttering severity and number of sessions to achieve discharge criteria).
Results: One-half (50%) of participants exhibited elevated ADHD symptoms. These children required 25% more clinical intervention time to achieve successful fluency outcomes than children without elevated ADHD symptoms. Findings suggest that more ADHD symptoms, increased pre-treatment stuttering severity, and male sex were associated with poorer responsiveness to stuttering treatment.
Conclusion: The large proportion of children exhibiting elevated ADHD symptoms, and the increase in clinical contact time required in this subgroup to achieve successful fluency outcomes, is suggestive of the need for clinicians to tailor stuttering intervention to address these concomitant behaviour challenges. Findings support the use of careful caseload management strategies to account for individual differences between children, and strengthen prognostic information available to parents and clinicians.

1. Introduction

Contemporary multidimensional models of early stuttering highlight interactions between language, phonology, physiology, temperament and behaviour in children predisposed to the neurodevelopmental disorder of stuttering (Smith & Weber, 2016, 2017). Inherent in such models is the importance of early intervention given the importance of neuroplasticity (Chang, Erickson, Ambrose, Hasegawa-Johnson, & Ludlow, 2008), and the importance of considering individual factors that may affect treatment outcomes. While successful early stuttering interventions have been reported (De Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015; Guitar

et al., 2015; Millard, Nicholas, & Cook, 2008; Yaruss, Coleman, & Hammer, 2006), authors caution that not all therapies work for every child (Smith & Weber, 2016; Yaruss et al., 2006). This was the case, for example, in the recent RESTART randomised controlled clinical trial comparing the efficacy of direct and indirect stuttering therapies (De Sonneville-Koedoot et al., 2015). An average of 26% of children who received treatment (46 out of the 176) across both therapy approaches still required therapy at 18 months post treatment, indicating that the disorder had not yet been fully remediated. Type of treatment (i.e., direct or indirect), as well as interactions between treatment type and time in therapy, age, stuttering severity and time close to onset, were examined and found not to be predictive of treatment outcomes (De Sonneville-Koedoot et al., 2015). This underscores the need to consider additional factors to achieve positive treatment outcomes for children who do not respond to stuttering therapy.

1.1. Temperament and self-regulation in children who stutter

One factor that has received considerable attention in stuttering research is the temperament characteristics of the child. Particular aspects of temperament known to influence stuttering include emotional reactivity and self-regulation (Arnold, Conture, Key, & Walden, 2011; Eggers, De Nil, & Van Den Bergh, 2013; Jones, Conture, & Walden, 2014; Karrass et al., 2006). Emotional reactivity refers to how an individual responds to changes in the environment, and self-regulation refers to the modulation or control of such reactivity (Onchwari & Keengwe, 2011). Research has found that children who stutter have significantly increased emotional reactivity and poorer self-regulation skills compared to normally fluent children (Karrass et al., 2006). Children with poor self-regulation are more likely to exhibit intense emotions, have difficulty calming when upset, and experience challenges with focusing and shifting attention between activities (Posner & Rothbart, 2009). Self-regulation is of particular relevance to early stuttering because the child exercises self-regulation in order to inhibit the stuttering behaviour and to attend to therapy strategies within and beyond clinic (Eggers et al., 2013).

Previous research has suggested that the temperament construct of effortful control is correlated with stuttering severity among children who stutter (Kraft, Ambrose, & Chon, 2014). Kraft et al. (2014) examined a range of factors that may be predictive of stuttering severity, including age, sex, socio-economic status, negative life events, home environment and temperament. Of all factors tested, the only factor found to be predictive of stuttering severity was the temperament construct of effortful control as measured in the Child Behaviour Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001). In the CBQ, effortful control is reflected in four subscales, which together form the key components of self-regulation (Backer-Grøndahl, Nærde, Ulleberg, & Janson, 2016): (a) inhibitory control, referring to "the capacity to suppress a dominant response and to plan actions" (p. 101); (b) attention focusing, referring to "the ability to concentrate and focus attention" (p.101); (c) low intensity pleasure, referring to "enjoyment of low-intensity activities" (p. 101); and (d) low perceptual sensitivity referring to the "awareness of and sensitivity to low-intensity stimulation in the environment" (p. 101). Kraft and her colleagues' (2014) results were replicated with a larger sample by Kraft, Beilby and Lowther (2016). In both studies, a significant negative relationship was found between both clinician and parent-reported stuttering severity ratings and effortful control, suggesting that children with more severe stuttered speech exhibited significantly lower effortful control.

Reduced self-regulation is highlighted in the Demands and Capacities Model as a potentially significant component in the development of stuttering (DCM; Starkweather, 1987). According to the DCM, stuttering arises and is maintained by an imbalance between demands, that is, external environmental pressures including cognitive, linguistic, motor, and socio-emotional factors, and capacities, that is, the speech motor planning and execution abilities or skills required for fluent speech (Starkweather, 1987). Children who stutter with poor ability to self-regulate are more likely to respond with increased volatility, for example, by over-reacting or reacting unpredictably to changes in their environment. The DCM theory suggests that such emotional reactions may place increased motoric and emotional demands on the speech motor system, subsequently reducing the capacity for controlled fluent speech production (Starkweather, 1987). Furthermore, these children may have greater difficulty with allocation of attentional resources, possibly reducing the ability to allocate sufficient attention to therapy strategies and to the production of fluent speech (Eggers et al., 2013).

1.2. Self-regulation and attention deficit hyperactive disorder (ADHD)

Challenges in self-regulation for children who stutter (Karrass et al., 2006) appear similar to those experienced by some children with ADHD symptoms, given that a core challenge for children with ADHD is emotional dysregulation (Barkley, 2013). Self-regulation deficits in children who stutter have been demonstrated in poorer attentional shifting and focusing of control (Eggers, De Nil, & Van de Bergh, 2010, 2012; Karrass et al., 2006). In addition, behaviours such as more frequent daydreaming (Alm & Risberg, 2007) and an overall lower perceptual sensitivity to sensory input from the environment also distinguish children who stutter from their fluent peers (Eggers, De Nil, & Van den Bergh, 2010; Embrechts, Ebben, Franke, & Van de Poel, 2000).

Self-regulation is of particular interest in early stuttering given the need the child has to exercise self-regulation in order to inhibit the stuttering behaviours and to attend to therapy strategies within and beyond clinic (Arnold et al., 2011; Karrass et al., 2006). Furthermore, poor self-regulation skills are predictive of long-term negative emotional, social, and occupational outcomes (Moffitt et al., 2011; Wiersema & Roeyers, 2009). Eggers, De Nil, and Van Den Bergh (2012) compared the attentional networks in children who stutter and children who do not stutter using the Attention Network Test. The role of attentional networks in self-regulation can be traced back to early infancy, as infants as young as 3 months old develop the ability to orient to a familiar visual stimulus for soothing and distress reduction (Posner & Rothbart, 2009). Children who stutter were found to exhibit lower efficiency in the orienting attention network, pointing to "a possible role for attentional processes in developmental stuttering" (Eggers et al., 2012, p.

956). Findings were taken to suggest that children who stutter might be less able to orient their attentional resources between concurrent tasks, including speech planning and production. This is consistent with the DCM, which highlights the impact of environmental demands on attentional resource allocation in children who stutter (Starkweather, 1987). Children who stutter with greater attentional allocation challenges may have greater difficulty allocating resources to fluency strategies and speech motor control, within and beyond-clinic, than those with more efficient attention networks.

Poor self-regulation skills have been found to be prevalent in children who stutter, and the recent study by Kraft et al. (2016) highlights that children with more severe stuttered speech have greater challenges in exerting effortful control over their behaviour in general (Alm & Risberg, 2007; Choi, Conture, Walden, Lambert, & Tumanova, 2013; Eggers et al., 2010; Jones et al., 2014; Karrass et al., 2006; Zengin-Bolatkale, Conture, Key, Walden, & Jones, 2018). The proportion of children who stutter presenting with self-regulation difficulties, resembling those seen in ADHD, has not been rigorously investigated and remains of significant interest for potential management in clinical practice; as does the possible effect of elevated ADHD symptomology on treatment responsiveness.

Previous studies have examined the prevalence of ADHD symptoms in children who stutter at a subclinical level. Such studies are small in number and are difficult to interpret due to relatively limited sample sizes (Donaher & Richels, 2012; Riley & Riley, 2000). In a clinical sample of 36 children, Donaher and Richels (2012) identified 58% of children who stutter as having subclinical ADHD symptoms. In a sample of 50 children, Riley and Riley (2000) identified 26% of children who stutter as presenting with a concomitant attending disorder. It was not evident if the "attending disorder" reflected subclinical symptoms or a clinical diagnosis.

Other studies have not directly quantified the proportion of children who stutter with subclinical ADHD symptoms, though they have found that a significantly greater proportion of these children exceed the threshold of clinical concern for ADHD symptoms (Alm & Risberg, 2007; Felsenfeld, Van Beljstervveldt, & Boomsma, 2010). A large-scale longitudinal study of 20,445 twin children suggested the possible existence of two subgroups of children who stutter: one group with concomitant attentional deficits and another group without such attentional problems (Felsenfeld et al., 2010). The ADHD symptoms in the aforementioned studies did not necessarily mean that the diagnostic criteria for ADHD were met. Nevertheless, the authors indicated that predominantly subclinical ADHD symptoms may contribute to the onset and expression of developmental stuttering, and that these symptoms should be considered in management of this complex disorder (Felsenfeld et al., 2010).

The prevalence of ADHD in children who stutter is unclear. Blood, Ridenour, Qualls, and Hammer (2003) reported the prevalence to be 5.9%, only slightly higher than the prevalence of 3–5% in the normally fluent school age population (Anastopoulos & Shelton, 2001). Ultimately, clinical levels of ADHD symptoms may not be a key feature in early stuttering; however, at least subclinical symptoms of ADHD appear to occur in a large proportion of children who stutter. These children may require an integrated intervention addressing concomitant behaviors in addition to fluency to optimise treatment for terms of stuttering as well as inattentiveness, hyperactivity and impulsivity.

1.3. The present study

Previous research has highlighted the importance of temperament in children who stutter, with particular focus on the possible role of self-regulation and attentional challenges on stuttering severity and treatment success (Kraft et al., 2014; Riley & Riley, 2000). However, no previous research has attempted to quantify the effect of these self-regulation challenges, resembling ADHD-like symptoms, on treatment responsiveness, while controlling for the effect of other clinical variables of interest. Furthermore, the proportion of children who stutter presenting to clinic with these elevated ADHD-like symptoms warrants further investigation, investigated in only one previous study with a relatively small sample size (Donaher & Richels, 2012).

This study aimed to determine the proportion of children who stutter who also exhibit elevated ADHD symptomology. This is similar to the approach in Donaher and Richels (2012) but applied to a large sample size. ADHD symptoms were measured by the ADHD Rating Scale (McGoey, DuPaul, Haley, & Shelton, 2007; Power, Costigan, Leff, Eiraldi, & Landau, 2001), a parent-report measure that has been widely used to screen for ADHD in both research and clinical practice. Consistent with Donaher and Richels (2012), children were classified as exhibiting elevated ADHD symptoms if they scored a rating of 2 (often) or greater on any 2 items related to inattention (inattentive type), or any 2 items related to hyperactivity/impulsivity (hyperactive/impulsive type; HI). In the current study, the participant's overall score on the scale also had to be equal to or greater than the 80th percentile for their age and sex (McGoey et al., 2007; Power et al., 2001).

A retrospective clinical audit research design was employed whereby participants' demographic, pre-treated stuttered speech, and treatment data were obtained from clinical data records from a combination of public and private clinics. An evaluation of the clinical data comparing children who stutter with elevated ADHD symptoms to those without these concomitant behaviours on the following demographic variables (age at onset, time between onset of stuttering and therapy commencement, family history of developmental stuttering disorder and sex), clinical stuttered speech behaviours (pre-treatment stuttering severity, and pretreatment typography), and treatment data (post-treatment stuttering severity and number of treatment sessions) was conducted.

This also provided the opportunity undertake a preliminary exploration of the effect of elevated ADHD symptoms on stuttering therapy outcomes. Stuttering therapy outcomes were measured in terms of the number of therapy sessions required to achieve normal fluency. Consenting participants' data were still included in the analyses if they did not achieve normal fluency. It was predicted that the prevalence of elevated ADHD symptoms amongst children who stutter would be approximately 50–60% (Donaher & Richels, 2012). Further, it was predicted that the presence of elevated levels of ADHD symptoms would increase the required number of therapy sessions, as well as explain unique variance in treatment responsiveness Riley & Riley, 2000).

Data were also collected on a number of stuttering therapy variables that have received attention in the stuttering therapy literature but continue to be debated in terms of treatment responsiveness (Clark, Tumanova, & Choi, 2017). These included age at

onset of stuttered speech, time from onset to the start of treatment, family history, sex, stuttering severity and disfluency types (typography). Such variables were included as covariates in the analysis, if identified as significantly correlating with the outcome measure of treatment responsiveness.

2. Methods

2.1. Participants

Participants were families of children with clinically diagnosed stuttering, recruited from the *Author De-Identified* Stuttering Treatment Clinic (CSTC) and four specialised community clinics in the metropolitan area of Perth, Western Australia, all within 20 km of the city center. Caseloads comprised children from metro, rural and country locations up to 460 km from the city center. Thirty-four percent of the sample was recruited from CSTC and the remainder recruited from community clinics (with an approximately equal proportion recruited from each clinic).

The sample comprised participants who had completed and been discharged (or discontinued) from stuttering therapy within the last three months prior to onset of the study. Discharge criteria were the same across all clinics and included a within clinic rating of stuttering severity ≤ 2% syllables stuttered, as well as a rating at or below 2, on caregiver perceptual rating scales over a period of three months (Manning, 2010; Yairi & Ambrose, 1999). Two hundred and seven participants were contacted, and 185 responded. The mean age of participants at the start of therapy was 3 years 5 months (range: 2 years, 2 months to 6 years, 3 months).

In 15 cases (8.1%), the families of participants terminated therapy despite not meeting formal discharge criteria. Of these 15 cases, 12 participants were resistant to therapy techniques, two withdrew secondary to health issues and one family moved interstate. Furthermore, 11 of these 15 participants (73%) were classified as having elevated symptoms of ADHD and four (27%) did not meet criteria for elevated ADHD symptoms, pointing to a possible role of these symptoms in resistance to treatment. These participants were included in the study to strengthen clinical validity of findings in the professional community, reflecting realistic clinical outcomes where attrition of clients may occur before discharge criteria is met. An analysis excluding these participants, however, did not change the results regarding predictors of treatment responsiveness.

2.2. Measures

The ADHD Rating Scale- IV Home Version (McGoey et al., 2007; Power et al., 2001) is a parent-completed, 18-item questionnaire directly adapted from the ADHD symptom lists as specified in the DSM-V diagnostic criteria (American Psychological Association, 2013). It is a screening tool for ADHD and a measure of attention and child disruptive behaviour (Power et al., 2001). Factor analysis has indicated that the ADHD Rating Scale-IV comprises 9 items measuring inattention and 9 items measuring hyperactivity/impulsivity (DuPaul et al., 1998). Each item is rated by the parent on a 4-point scale, ranging from 0 (never or rarely), 1 (sometimes), 2 (often) to 3 (very often). The overall scale score ranges from 0 to 54, with higher scores indicating more ADHD symptoms. The scale has demonstrated strong psychometric properties, including concurrent validity, test-retest reliability (correlations ranging from .80 to .87) and internal consistency (DuPaul et al., 1998; McGoey et al., 2007). Two versions of the scale were used, one version for participants aged 2 to 4 years, 11 months (McGoey et al., 2007), and one version for participants from 5 to 6 years, 11 months (Power et al., 2001).

The scale was used to allocate children into two groups for between-group analyses: children who stutter who present with elevated ADHD symptoms, and those children who stutter who do not meet the criteria for elevated ADHD symptoms. Parents completed this scale at time of recruitment.

All participants completed a detailed pre-treatment case history as part of the initial assessment, including details such as age, sex and family history (Guitar, 2014). In addition, measures pertaining to stuttering severity and typography are entered on a weekly basis after each treatment session, as part of clinic treatment protocol (Guitar, 2014). All clinics followed the same protocol for data collection.

All clinical variables of interest (age at onset, time between onset of stuttering and therapy commencement, family history, sex, pre-treatment stuttering severity and typography, number of treatment sessions, post treatment stuttering severity) were obtained from client database records, comprising regular clinical progress entries following each client visit. Age at onset was measured in years and months, time between onset of stuttering and therapy commencement was measured in weeks, and family history of stuttering was coded as a categorical variable. Pre and post-treatment stuttering severity were measured by percentage of syllables stuttered, calculated by the Fluency Rater Application (ForFluency, 2012). Severity ratings were made on within-clinic conversations with the clinician using a minimum of 500 syllables. Disfluencies included syllable, part-word and monosyllabic-word repetitions, sound prolongations, and both inaudible and audible blocks (i.e. the stoppage of airflow in the initiation of speech) (Guitar, 2014; Smith & Weber, 2017). Stuttering typographies were coded as three separate binary variables reflecting the presence or absence of predominantly repetitions (sounds, syllables or one-syllable words), sound prolongations, and blocks (audible or inaudible) at pre-treatment assessment (Guitar, 2014; Smith & Weber, 2017).

The outcome variable of "number of sessions" was also obtained from client database records. Ninety two percent (170/185) of participants in the present study reached the discharge criteria. There was no significant difference between groups of children who stutter with and without ADHD symptoms in their post-treatment stuttering severity ratings. As such, the post-treatment stuttering severity score was not of interest for this study. Instead, the possible impact of elevated ADHD symptomology on time taken to achieve discharge criteria was examined, based on the number of clinical therapy sessions required to achieve the discharge criteria.

2.3. Procedure

Clinic managers from the private speech pathology stuttering treatment clinics contacted all families of children who had been discharged from therapy within the three months prior to the start of this study, via standard mail or email, and asked for their interest in participating in the project. Participants who responded positively were sent information sheets and informed consent forms, as well as an electronic version of the ADHD Rating Scale (total completion time 5–8 minutes) using Qualtrics Survey Software. The Scale was renamed the Behaviour Profile Scale to eliminate stigma attached to ADHD diagnoses and possible misreporting (DosReis, Barksdale, Sherman, Maloney, & Charach, 2010). Parents were instructed to complete the consent form and fill in the Behaviour Scale electronically. Once consent was obtained, clinic managers accessed participant data from the clinic management software or hard copy clinical progress notes on key variables of interest for the study. The study was approved by the Curtin University Human Research Ethics Committee

2.4. Data analysis

Firstly, the analysis identified the proportion of children who stutter who presented with elevated ADHD symptomology in relation to the total number of participants. Secondly, because of violations to the assumption of normality, non-parametric Mann Whitney U tests were used to test for differences between those children with elevated ADHD symptomatology and those without on all continuous variables, and chi-square tests of contingencies were used to compare groups in terms of family history, sex and stuttering typography.

Finally, generalized linear mixed modelling (GLMM) was used to examine whether elevated ADHD symptomology uniquely predicted treatment response (i.e., number of therapy sessions) while controlling for potential covariates. GLMM is robust to assumption violations (e.g., non-normal distributions) and can manage mixed categorical and continuous variables; it can also handle missing data without excluding participants (Kain, Bolker, & McCoy, 2015). Spearman's *rho* correlations were conducted to identify if any potential covariates were correlated significantly with the outcome variable; if they were, they were included as covariates in the GLMM. Unstandardised and standardized regression coefficients, as well as squared semi-partial correlations, were obtained from the corresponding standard multiple regression, and *p*-values were obtained from the GLMM.

3. Results

The proportion of children who stutter with elevated ADHD symptomology out of the total sample was found to be 49.73% (92 out of the 185 participants). Further examination of individual ADHD subscales was carried out, and the proportion of children scoring at or above the 80th percentile for inattentive or HI symptoms were identified. Of the 92 participants who met criteria for elevated ADHD symptomology, 66 also met criteria for elevated levels of inattention only and 67 also met criteria for HI only. Seven participants met criteria for inattention who did not also meet criteria for elevated combined ADHD symptoms. Five participants met criteria for HI who did not also meet criteria for elevated combined ADHD symptoms.

Based on the ADHD parent scale, 92 children who stutter in the final sample were identified as having elevated symptoms of ADHD. See Tables 1 and 2 for participant characteristics of those with and without elevated ADHD symptoms, and Table 2 for between group differences in binary variables. According to Mann-Whitney U tests, the only variable found to be significantly different between groups was number of treatment sessions, U = 3440.00 (p = .02). Specifically a 24.06% increase in total number of clinical visits (approximately 3 sessions) was required to achieve discharge criteria in the group of children with elevated ADHD symptoms. Between-group differences in age at onset and time from onset to start of treatment and pre-treatment stuttering severity were negligible. Although the between-group difference in post treatment stuttering severity did not reach significance, the mean of 1.60%SS in the group of children without elevated ADHD symptoms meets criteria for fluency within normal limits. The mean of 2.24%SS in the group of children with elevated ADHD symptoms indicates that the average stuttering severity at post treatment was slightly above the discharge criteria for remediation of the disorder (i.e. ≤ 2%SS).

Chi-square tests of contingencies were conducted to examine the relationship between group membership and categorical variables (family history, sex and stuttering typography). No significant relationships were found between group membership and all

Table 1
Participant demographics (categorical variables).

Variable	CWS only Number/93 (%)	CWS + eADHD Number/92 (%)
Sex (male)	56 (60.2)	63 (68.5)
Sex (female)	37 (39.8)	29 (31.5)
Family history	73 (78.5)	80 (87.0)
Typography (repetitions)	77 (82.8)	75 (81.5)
Typography (prolongations)	23 (24.7)	17 (18.5)
Typography (blocks)	23 (24.7)	23 (24.7)

Note. CWS only = group of children who stutter without elevated ADHD symptoms. CWS + eADHD = group of children who stutter with elevated ADHD symptoms.

Table 2

Between group differences in stuttering therapy variables using Mann Whitney U tests.

Variable	CWS only $n = 93$		CWS + eADHD $n = 92$		Z	p	Cohen's r
	M	SD	M	SD			
Age at onset (years)	2.76	0.79	2.90	0.80	− .94	.35	− .07
Time close to onset (weeks)	28.88	36.38	31.80	35.84	− .70	.49	− .05
Pre-treatment %SS	15.49	7.23	15.82	6.68	− .47	.65	− .04
Post-treatment %SS	1.60	0.95	2.24	2.81	− 1.14	.26	− .08
Number of sessions	11.16	5.33	13.80	7.57	− 2.31	.02	.17

Note. CWS only = group of children who stutter without elevated ADHD symptoms. CWS + eADHD = group of children who stutter with elevated ADHD symptoms.
%SS refers to percentage of syllables stuttered.

three categorical variables tested ($p > .05$) (see Table 3).

Bivariate and point biserial Pearson correlations were run to determine the covariates to be included in the GLMM analysis (see Table 4). If variables were found to be significantly correlated with "number of sessions" they were included in the GLMM. Both male sex and increased stuttering severity were associated with an increased number of sessions required to achieve successful therapy outcomes. The negative correlation between the stuttered speech behavior typography of repetitions and number of treatment sessions shows a trend for children who present with predominantly repetition type stutters (vs. those who do not) to have required fewer therapy sessions. On average, male participants received 13.48 clinical intervention sessions, while females received 10.65 sessions. Participants with predominantly repetitions-type stutters required 11.98 sessions to achieve discharge criteria, while participants without predominantly repetition-type stutters at pre-treatment (prolongation and/or block type stutters only) required an average of 14.73 therapy sessions. As such, sex, pre-treatment stuttering severity and repetition type stutters were included as covariates in the GLMM. The correlations between prolongations and block-type stutters showed a trend for children without more advanced type stutters to require more therapy sessions, but were not statistically significant and were therefore not entered into the model.

GLMM was conducted to explore if elevated total ADHD symptoms, as well as separate inattentive symptoms and hyperactive/impulsive symptoms, were predictive of responsiveness to stuttering treatment (while controlling for previously identified covariates, see Table 5). As such, three separate GLMM analyses were conducted to identify the possible differences in variance accounted for when the ADHD combined scale score was divided into its components.

Table 3

Relationships between group membership and binary variables.

	1. ADHD			2. Inattention			3. HI			4. FamHx			5. Sex			6. Repetitions			7. Prolongations			8. Blocks
	X^2	p	φ	X^2	p	φ	X^2	p	φ	X^2	p	φ	X^2	p	φ	X^2	p	φ	X^2	p	φ	
2.	79.8[a]	≤ .001	.66																			
3.	88.5[b]	≤ .001	.69	53.0[c]	≤ .001	.54																
4.	2.3	.13	.11	3.4	.07	.14	.03	1.0	.01													
5.	1.4	.24	.09	4.9[d]	.03	.16	5.85[e]	.02	.18	.96	.33	.07										
6.	.1	.82	.02	.2	.70	.03	.72	.34	.43	3.6	.08	.14	.51	.48	.05							
7.	1.1	.30	.08	1.0	.31	.08	.03	.43	.06	2.1	.15	.11	.22	.64	.04	13.5[f]	≤ .001	.27				
8.	< .01	.97	< .01	.09	.77	.02	.54	.49	.05	.2	.64	.03	.25	.62	.04	62.5[g]	≤ .001	.58	8.5[h]	.004	.21	

Note.

HI refers to ADHD hyperactive/impulsive symptoms. FamHx refers to family history of stuttering.

[a] Out of 92 participants who met criteria for ADHD combined, 66 also met criteria for inattention; out of 93 participants who did not meet criteria for ADHD combined, 7 met criteria for inattention.

[b] Out of 92 participants who met criteria for ADHD combined, 67 also met criteria for HI; out of 93 participants who did not meet criteria for ADHD combined, 5 met criteria for HI.

[c] Out of 73 participants who met criteria for inattention, 52 also met criterion for HI; out of 112 who did not meet criteria for inattention, 20 met criteria for HI.

[d] Out of 73 participants who met criteria for inattention, 54 were male and 19 were female; out of 112 who did not meet criteria for inattention, 65 were male and 47 were female.

[e] Out of 72 participants who met criteria for HI, 54 were male and 18 were female; out of the 113 who did not meet criteria for HI, 65 were male and 48 were female.

[f] Out of 40 participants who presented with prolongations, 25 also presented with repetitions; out of 145 participants who did not present with prolongations, 127 presented with repetitions.

[g] Out of 152 participants who presented with repetitions, 20 also presented with blocks; out of 33 who did not present with repetitions, 26 presented with blocks.

[h] Out of 40 participants who presented with prolongations, 17 also presented with blocks; out of 145 participants who did not present with prolongations, 29 presented with blocks.

Table 4
Correlation matrix.

	ADHD	IA	HI	%SS	Age	TTO	NoSx	FamHx	Sex	Rep	Prolong	Block
ADHD	1	.964**	.961**	.16*	.11	.08	.27**	.06	−.17*	−.03	−.04	.04
IA	.964**	1	.86**	.15*	.12	.07	.3**	.09	−.15*	−.01	−.03	.02
HI	.961**	.86**	1	.17*	.09	.09	.22**	.01	−.17*	−.05	−.05	.07
%SS	.16*	.15*	.17*	1	.03	.07	.26**	.16*	−.1	−.38**	.30**	.28**
Age	.11	.12	.09	.03	1	−.05	.08	−.09	.01	.19	−.20**	−.09
TTO	.08	.07	.09	.07	−.05	1	−.05	−.18*	.02	−.08	.08	.04
NoSx	.27**	.30**	.22**	.26**	.08	−.05	1	.1	−.20 a **	−.16 b*	.12	.13
FamHx	.06	.09	.01	.16*	−.09	−.18*	.1					
Sex	−.17*	−.15*	−.17*	−.11	.01	.02	−.20 a **					
Rep	−.03	−.01	−.05	−.38**	.19	−.08	−.16 b*					
Prolong	−.04	−.03	−.05	.30**	−.20**	.08	.12					
Block	.04	.02	.07	.28**	−.09	.04	.13					

Note: ** Correlation is significant at the .01 level. * Correlation is significant at the .05 level. ADHD = ADHD combined score (combined total of IA and HI scores). IA = ADHD inattentive symptoms. HI = ADHD hyperactive/impulsive symptoms. %SS = percentage of syllables stuttered at pre-treatment. Age = age at onset of developmental stuttering disorder. TTO = weeks between onset of developmental stuttering disorder and commencement of treatment. NoSx = number of therapy sessions. FamHx = family history of stuttering. Rep = repetition type disfluencies. Prolong = prolongation type disfluencies. Block = block type disfluencies.

Table 5
Generalised linear mixed model predicting number of sessions from covariates: pre-treatment stuttering severity, sex and repetitions type disfluencies, with ADHD combined scores, Inattention and Hyperactivity/Impulsivity scores as primary predictors.

Predictors (IVs)	B	95% CI	β	sr^2	p-value[1]
Dependant Variable: Number of Sessions					
Step 1					
PreSS	.203	.06, .35	.21	.04	.001
Sex	−2.47	−4.40, -.54	−.18	.034	.003
Rep	−1.18	−3.79, 1.42	−.07	.004	.367
$F(3, 181) = 6.85, p < .001$					
$R^2 = .102$					
Primary predictor: ADHD					
Step 2					
PreSS	.17	.03, .31	.18	.03	.003
Sex	−2.02	−3.94, -.11	−.15	.023	.016
Rep	−1.33	−3.88, 1.22	−.08	.006	.283
ADHD	.09	.03, .15	.21	.046	.004
ADHD: $\Delta F(1, 180) = 8.73, p = .004$ $F(4, 180) = 7.54, p < .001$					
$R^2 = .144$					
$\Delta R^2 = .042$					
Primary predictor: Inattention					
Step 2					
PreSS	.16	.02, .31	.17	.028	.005
Sex	−1.98	−3.87, -.09	−.14	.023	.023
Rep	−1.44	−3.97, 1.08	−.08	.007	.234
Inattention	.204	.09, .31	.26	.069	.002
Inattention: $\Delta F(1, 180) = 13.43$ $p < .001$ $F(4, 180) = 8.85, p < .001$					
$R^2 = .164$					
$\Delta R^2 = .062$					
Primary predictor: HI					
Step 2					
PreSS	.18	.04, .32	.19	.032	.002
Sex	−2.12	−4.06, -.18	−.15	.025	.010
Rep	−1.23	−3.81, 1.34	−.07	.005	.330
HypImp	.14	.02, .26	.16	.027	.015
HI: $\Delta F(1, 180) = 5.001, p = .027$					
$R^2 = .126$					
$\Delta R^2 = .024$					

Note.[1]GLMM adjusted values. B = unstandardized regression coefficients. β = standardised regression coefficients. sr^2 = squared semi-partial correlations. PreSS = pre-treatment stuttering severity. rep = repetition type disfluencies. ADHD = combined inattention and hyperactive/impulsive symptoms. Inattention = ADHD inattentive symptoms. HI = ADHD hyperactive/impulsive symptoms.

On Step 1 of the hierarchical multiple linear regression, pre-treatment stuttering severity, sex and repetition type stutters accounted for a significant 10.2% of the variance in treatment responsiveness (number of sessions). When the combined ADHD score was the primary predictor, ADHD total scores were added to the regression equation on step 2, and accounted for an additional 4.2%

of the variance in treatment responsiveness. In combination, the four predictor variables explained 14.4% of the variance in treatment responsiveness.

When the inattentive subscale scores was the primary predictor, inattentive scores were added to the regression equation on step 2, and accounted for an additional 6.2% of the variance in treatment responsiveness. In combination, the four predictor variables explained 16.4% of the variance in treatment responsiveness.

When the individual HI subscale scores was the primary predictor, HI scores were added to the regression equation on step 2, and accounted for an additional 2.4% of the variance in treatment responsiveness. In combination, the four predictor variables explained 12.6% of the variance in treatment responsiveness.

In light of the slightly greater variance in treatment responsiveness accounted for by inattention symptoms, when compared to combined symptoms and HI symptoms, partial correlations were conducted to further examine the relationship between the two individual subscales and number of sessions. A significant relationship was found between inattention and number of sessions while controlling for HI symptoms, $r(182) = 0.23$, $p = .002$. A non-significant relationship was found to exist between HI symptoms and number of sessions while controlling for inattention, $r(182) = -.08$, $p = .28$. This preliminary finding may suggest a possibly greater role of attentional challenges in treatment responsiveness.

4. Discussion

Contemporary stuttering research has highlighted the importance of the temperament characteristics—and, in particular, self-regulation challenges—of children who stutter. In this light, the overall poorer self-regulation skills (Karrass et al., 2006) characteristic of elevated ADHD symptomology warrant investigation in this disorder (Felsenfeld et al., 2010; In addition, assumptions have been made about the possible impact of attentional difficulties on therapy success (Riley & Riley, 2000) and about the possibility of the shared etiological roots of stuttering and attentional challenges (Felsenfeld et al., 2010). Furthermore, Riley and Riley (2000) found that children who stutter with attentional deficits were less successful in maintaining positive outcomes in stuttering therapy if their attentional deficits were not addressed. However, little research has attempted to quantify the effect of these ADHD-like symptoms on treatment responsiveness. This study provides preliminary evidence of the impact of poor self-regulation skills (characterised by inattentive and HI symptoms) on early stuttering treatment responsiveness, while evaluating the possible effect of other noteworthy clinical variables.

Approximately half of the children who stutter (49.73%) in the present study presented with concomitant elevated ADHD symptoms. This is in line with research identifying elevated ADHD symptomology in children who stutter compared to their fluent peers (Alm & Risberg, 2007; Donaher & Richels, 2012; Felsenfeld et al., 2010), and similar to the proportion reported by Donaher and Richels (2012). Early intervention stuttering treatment relies heavily on successful parent implementation of therapy contingencies and strategies beyond the clinic (Millard et al., 2008). The large proportion of children who stutter exhibiting elevated ADHD symptoms underscores the responsibility that clinicians have to support parents to address these concomitant behavioural challenges in their children, as self-regulation skills can be successfully improved by parent behavioural intervention (Moffitt et al., 2011; Sanders & Mazzucchelli, 2013; Sanders, Kirby, Tellegen, & Day, 2014). Addressing these symptoms is crucial as self-regulation skills have been found to predict physical health, substance dependence, personal finances and criminal offending outcomes at 32 years of age (Moffitt et al., 2011).

Children with poorer ability to orient attention are less able to regulate their emotions and arousal in both positive (e.g., counting down to a family holiday) and negative situations (e.g., fighting with a sibling about a toy) (Rueda, Posner, & Rothbart, 2005). As a result of the reduced skills in attentional shifting in children who stutter (specifically those meeting criteria for "elevated ADHD inattentive symptoms" in the present study), children may be less able to shift their focus from high arousal situations by disengaging from emotion triggering stimuli (Rueda et al., 2005), thus placing increasing motoric and emotional demands on their speech motor systems (Starkweather, 1987). This may go some way in explaining the slightly greater variance in treatment responsiveness predicted by items on the ADHD Scale measuring inattention, as attentional challenges may affect resource allocation as well as distress modulation and emotional reactivity.

There were no significant differences found between the groups of children who stutter with and without elevated ADHD symptoms on demographic variables and pre-treatment stuttering features. In the present study, significant correlations were found between pre-treatment stuttering severity and ADHD symptoms, although correlations were weak. Relationships between lower effortful control and increased ADHD symptoms have been well documented (Wiersema & Roeyers, 2009; Samyn, Roeyers, & Bijttebier, 2011) and the correlations found in this study are consistent with those which previously found less effortful control to be correlated with increased stuttering severity (Kraft et al., 2014, 2018). Further research is recommended to explore both effortful control and ADHD symptoms in the context of early childhood stuttering to investigate this relationship further.

Children who stutter with elevated ADHD symptoms were found to require a greater number of therapy sessions to achieve discharge criteria. One possible implication that follows from this is that addressing the ADHD symptomatology in conjunction with stuttering therapy could lead to a decrease in the number of therapy sessions a child will need before discharge. Previous research has found children who stutter tend to have challenges across both sets of ADHD symptoms (Eggers et al., 2010, 2012; Eggers et al., 2013; Jones et al., 2014; Kraft et al., 2014, 2018). Similarly, in the present study both inattention and HI symptoms predicted treatment responsiveness, when considered in isolation. The inattentive symptoms, in particular, were most highly associated with treatment responsiveness and contributed unique variance, although not a substantive amount, to such treatment responsiveness, when controlling for the effect of HI. Smith and Weber (2017) declared that a more thorough understanding of speech behaviours in stuttering will leads to new or improved treatment protocols. Such protocols may include individualised components that address attention

issues in order to support therapeutic success.

A number of key variables of clinical interest were included in the analysis. Male sex was found to be significantly correlated with and predictive of poorer response to stuttering therapy. This poses challenges to previous research indicating sex not to be predictive of treatment response. Still, it supports findings that females are more responsive to therapy (Kingston, Huber, Onslow, Jones, & Packman, 2003; Rousseau, Packman, Onslow, Harrison, & Jones, 2007). Pre-treatment stuttering severity was found to be correlated with and predictive of treatment responsiveness. This is in line with findings by Guitar et al. (2015), who found pre-treatment stuttering speech severity to predict increased treatment duration, possibly reflective of reduced treatment responsiveness. A significant negative correlation was found to exist between the presence of a less advanced typography (predominantly repetition type stutter) and number of sessions, indicating that children with repetitive sounds, syllables or single words responded better to treatment support. Previous research has not directly examined stuttering typography as a predictor of treatment response, however research regarding the effect of typography on stuttering persistence is equivocal. Findings may offer preliminary support for research by Yairi and Ambrose (1999), who found more advanced typography (e.g., prolongations and block type stutters) to be strongly correlated with persistent stuttering. However, other authors have reported no significant difference in the presence of more advanced typography in the persistence of stuttering (Throneburg & Yairi, 2001). Future research may examine stuttering typography in the context of treatment response, as well as stuttering persistence versus recovery.

4.1. Strengths and limitations

The present study was a relatively large-scale clinical audit addressing an area of limited research to date that has relevance for clinicians charged with managing challenging concomitant behaviors in children who stutter. A number of limitations were identified in the present study. All five clinics from which participants were recruited were speech pathology clinics specialising in the management of early childhood stuttering. As such, the proportion of children who stutter with elevated ADHD symptoms may be higher than children presenting to general community speech pathology clinics. Furthermore, the commitment by caregivers of these children may be greater and therefore they may be more motivated to facilitate responsiveness to change in these specialised clinical settings.

Given the retrospective nature of the study, "number of sessions" was a consistent measure of treatment responsiveness that was available in clinical records across all clinics from which participants were recruited. In spite of the number of treatment sessions to discharge being a relatively gross measure of treatment responsiveness, the present study has yielded some significant findings. Future prospective clinical research focusing on the possible effect of ADHD symptomology should, however, consider alternative approaches to measuring treatment responsiveness, such as monitoring changes in fluency after a fixed number of treatment sessions.

Future research is also needed before firm implications for clinical management of stuttering when a child has elevated symptoms of ADHD can be proposed. While treatment is indicated for children with clinically diagnosed ADHD, the potential benefit of treating subclinical ADHD symptoms in the context of stuttering therapy has yet to be established. The present findings justify undertaking such investigations.

Given that this study used retrospective clinical data, and that parents completed the ADHD scale prospectively within 3 months post treatment, measures may have been subject to additional variability than might be associated with a controlled, prospective clinical trial. Although this is a strength in terms of ecological validity, a more controlled prospective study may provide a more accurate estimate of the effect of elevated ADHD symptoms on treatment effectiveness.

Natural recovery was not accounted for in terms of treatment responsiveness. However, given the fact that the literature is equivocal in terms of predicting natural recovery, and the known natural recovery rate for preschool children sits at 6.3% at 12 months after onset (Reilly et al., 2013), clinics from which participants were recruited from encourage treatment as close to onset as possible. Finally, given the preliminary nature of the investigation into the role of ADHD symptoms on treatment responsiveness, detailed information regarding content of treatment sessions was deemed beyond the scope of the study. See Druker, Mazzucchelli, and Beilby (2018) for an outline of treatment conducted.

5. Conclusion

Empirical data is required to objectively capture the important prevalence of elevated ADHD symptoms in children with early developmental stuttering disorders. This study identified approximately half of the children in early stuttering caseloads as presenting with concomitant self-regulation challenges. Furthermore, this study explored the significant negative effect of elevated ADHD symptoms on stuttering treatment responsiveness and thus provides support for future prospective studies of treatments that address elevated ADHD symptomology as well as the child's stuttering. Finally, results yielded provide direction for future caseload and waitlist management, and potentially more informed individual prognostic information for parents of children who stutter who we treat in the future.

Conflict of interest

The authors report no conflicts of interest.

Funding

Acknowledgments

References

Alm, P. A., & Risberg, J. (2007). Stuttering in adults: The acoustic startle response, temperamental traits, and biological factors. *Journal of Communication Disorders, 40*(1), 1–41. https://doi.org/10.1016/j.jcomdis.2006.04.001.

Anastopoulos, A., & Shelton, T. (2001). *Assessing attention-deficit/hyperactivity disorders.* New York, NY: Kluwer Academic Publishers.

Arnold, H. S., Conture, E. G., Key, A. P., & Walden, T. (2011). Emotional reactivity, regulation and childhood stuttering: A behavioral and electrophysiological study. *Journal of Communication Disorders, 44*(3), 276–293.

American Psychological Association (2013). *American Psychiatric Association: Diagnostic and statistical manual of mental health disorders* (5th ed.). Arlington, VA: American Psychiatric Association.

Backer-Grøndahl, A., Nærde, A., Ulleberg, P., & Janson, H. (2016). Measuring effortful control using the children's behavior questionnaire-very short form: Modeling matters. *Journal of Personality Assessment, 98*(1), 100–109.

Barkley, R. (2013). *Taking charge of ADHD, third edition: The complete, authoritative guide for parents* (3th ed.). New York, NY: Guildford Publications.

Blood, G., Ridenour, V., Jr., Qualls, C., & Hammer, C. (2003). Co-occurring disorders in children who stutter. *Journal of Communication Disorders, 36*(6), 427–448.

Chang, S., Erickson, K., Ambrose, N., Hasegawa-Johnson, M., & Ludlow, C. (2008). Brain anatomy differences in childhood stuttering. *NeuroImage, 39*(3), 1333–1344. https://doi.org/10.1016/j.neuroimage.2007.09.067.

Choi, D., Conture, E., Walden, T., Lambert, W., & Tumanova, V. (2013). Behavioral inhibition in childhood stuttering. *Journal of Fluency Disorders, 38,* 171–183.

Clark, C., Tumanova, V., & Choi, D. (2017). Evidence-based multifactorial assessment of pre-school age children who stutter. *Perspectives of the ASHA Special Interest Groups, 2*(4), 4–27.

De Sonneville-Koedoot, C., Stolk, E., Rietveld, T., & Franken, M.-C. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PloS One, 10*(7), 1–17.

Donaher, J., & Richels, C. (2012). Traits of Attention Deficit/Hyperactivity Disorder in school-age children who stutter. *Journal of Fluency Disorders, 37*(4), 242–252.

DosReis, S., Barksdale, C., Sherman, A., Maloney, K., & Charach, A. (2010). Stigmatising experiences of parents of children with a new diagnosis of ADHD. *Psychiatric Services, 61*(8), 811–816.

Druker, K. C., Mazzucchelli, T. G., & Beilby, J. M. (2018). *An evaluation of an integrated fluency and resilience program for early developmental stuttering disorders.* Manuscript submitted for publication.

DuPaul, G. J., Anastopoulos, A. D., Power, T. J., Reid, R., Ikeda, M. J., & McGoey, K. E. (1998). Parent ratings of Attention-Deficit/Hyperactivity Disorder symptoms: Factor structure and normative data. *Journal of Psychopathology and Behavioral Assessment, 20,* 83–102.

Eggers, K., De Nil, L., & Van den Bergh, B. (2010). Temperament dimensions in stuttering and typically developing children. *Journal of Fluency Disorders, 35,* 355–372.

Eggers, K., De Nil, L., & Van Den Bergh, B. (2013). Inhibitory control in childhood stuttering. *Journal of Fluency Disorders, 38,* 1–13.

Eggers, K., De Nil, L. F., & Van Den Bergh, B. R. (2012). The efficiency of attentional networks in children who stutter. *Journal of Speech, Language and Hearing Research, 55*(3), 946–959.

Embrechts, M., Ebben, H., Franke, P., & Van de Poel, C. (2000). Temperament: A comparison between children who stutter and children who do not stutter. *Paper Presented at the Third World Congress on Fluency Disorders.*

Felsenfeld, S., Van Beljsterveldt, C., & Boomsma, D. I. (2010). Attentional regulation in young twins with probable stuttering, high nonfluency, and typical fluency. *Journal of Speech, Language, and Hearing Research, 53*(5), 1147–1166.

ForFluency (2012). *Fluency Rater. [Mobile application software].* Retrieved from http://download.cnet.com/Fluency-Rater/3000-2129_4-75357634.html.

Guitar, B. (2014). *Stuttering: An integrated approach to its nature and treatment* (4th ed.). Philadelphia, PA: Lippincott Williams & Wilkins.

Guitar, B., Kazenski, D., Howard, A., Cousins, S., Fader, E., & Haskell, P. (2015). Predicting treatment time and long-term outcomes of the Lidcombe Program: A replication and reanalysis. *American Journal of Speech-language Pathology, 24*(3), 533–544.

Jones, R., Conture, E., & Walden, T. A. (2014). Emotional reactivity and regulation associated with fluent and stuttered utterances of preschool-age children who stutter. *Journal of Communication Disorders, 48,* 38–51.

Kain, M., Bolker, B., & McCoy, M. (2015). A practical guide and power analysis for GLMMs: Detecting among treatment variation in random effects. *PeerJ, 3*(1226), 1–24.

Karrass, J., Walden, T., Conture, E., Graham, C., Arnold, H., Hartfield, K., et al. (2006). Relation of emotional reactivity and regulation to childhood stuttering. *Journal of Communication Disorders, 39,* 402–423.

Kingston, M., Huber, A., Onslow, M., Jones, M., & Packman, A. (2003). Predicting treatment time with the Lidcombe Program: Replication and meta-analysis. *International Journal of Language & Communication Disorders, 38*(2), 165–177.

Kraft, S. J., Ambrose, N., & Chon, H. (2014). Temperament and environmental contributions to stuttering severity in children: The role of effortful control. *Seminars in Speech and Language, 35*(2), 80–94.

Kraft, S. J., Beilby, J., & Lowther, E. (2016). The role of effortful control in stuttering severity in children: Australian replication study. Paper presented at American Speech and Hearing Association Convention. *Philadelphia.*

Manning, W. (2010). *Clinical decision making in fluency disorders.* San Diego, CA: Singular/Thomson Learning.

McGoey, K., DuPaul, G., Haley, E., & Shelton, T. (2007). Parent and teacher ratings of attention-deficit/hyperactivity disorder in preschool: The ADHD rating scale-IV preschool version. *Journal of Psychopathology and Behavioral Assessment, 29,* 269–276.

Millard, S., Nicholas, A., & Cook, F. (2008). Is parent-child interaction therapy effective in reducing stuttering? *Journal of Speech Language and Hearing Research, 51*(3), 636–650.

Moffitt, T., Arseneault, L., Belsky, D., Dickson, N., Hancox, R. J., Harrington, H., et al. (2011). Gradient of childhood self-control predicts health, wealth, and public safety. *Proceedings of the National Academy of Sciences of the United States of America, 108*(7), 2693–2698.

Onchwari, G., & Keengwe, J. (2011). Examining the relationship of children's behavior to emotion regulation ability. *Early Childhood Education Journal, 39,* 279–284.

Posner, M. I., & Rothbart, M. K. (2009). Toward a physical basis of attention and self-regulation. *Physics of Life Review, 6*(2).

Power, T. J., Costigan, T. E., Leff, S. S., Eiraldi, R. B., & Landau, S. (2001). Assessing ADHD across settings: Contributions of behavioral assessment to categorical decision making. *Journal of Clinical Child & Adolescent Psychology, 30*(3), 399–412.

Reilly, S., Onslow, M., Packman, A., Cini, E., Conway, L., Ukoumunne, O., et al. (2013). Natural history of stuttering to 4 years of age: A prospective community-based study. *Pediatrics, 132*(3), 460–467.

Riley, G., & Riley, J. (2000). A revised component model for diagnosing and treating children who stutter. *Contmeporary Issues in Communication Sciences and Disorders, 27*, 188–199.

Rothbart, K., Ahadi, S., Hershey, K., & Fisher, P. (2001). Investigations of temperament at three to seven years: The Children's Behavior Questionnaire. *Child Development, 72*(5), 1394–1408.

Rousseau, I., Packman, A., Onslow, M., Harrison, E., & Jones, M. (2007). An investigation of language and phonological development and the responsiveness of preschool age children to the Lidcombe program. *Journal of Communication Disorders, 40*(5), 382–397.

Rueda, R., Posner, M., & Rothbart, K. (2005). The development of executive attention: Contribution to the emergence of self regulation. *Developmental Neuropsychology, 28*, 573–594.

Samyn, V., Roeyers, H., & Bijttebier, P. (2011). Effortful control in typically developing boys and in boys with ADHD or autism spectrum disorder. *Research in Developmental Disabilities, 32*, 483–490. https://doi.org/10.1016/j.ridd.2010.12.038.

Sanders, M., Kirby, J., Tellegen, C., & Day, J. (2014). The Triple P-Positive Parenting Program: A systematic review and meta-analysis of a multi-level system of parenting support. *Clinical Psychology Review, 34*(4), 337–357.

Sanders, M. R., & Mazzucchelli, T. G. (2013). The promotion of self-regulation through parenting interventions. *Clinical Child and Family Psychology Review, 16*(1), 1–17.

Smith, A., & Weber, C. (2016). Childhood stuttering: Where are we and where are we going? *Seminars in Speech and Language, 37*(4), 291–297.

Smith, A., & Weber, C. (2017). How stuttering develops: The multifactorial dynamic pathways theory. *Journal of Speech Language and Hearing Research, 60*, 2483–2505.

Starkweather, W. C. (1987). *Fluency and stuttering.* Englewood Cliffs, N.J: Prentice-Hall.

Throneburg, R., & Yairi, E. (2001). Durational, proportionate and absolute frequency characteristics of dysfluencies: A longitudinal study regarding persistence and recovery. *Journal of Speech Language and Hearing Research, 44*(38–51).

Wiersema, J., & Roeyers, H. (2009). ERP correlates of effortful control in children with varying levels of ADHD symptoms. *Journal of Abnormal Child Psychology, 37*, 327–336.

Yairi, E., & Ambrose, N. (1999). Early childhood stuttering I: Persistence and recovery rates. *Journal of Speech Language and Hearing Research, 42*(5), 1097–1112.

Yaruss, J. S., Coleman, C., & Hammer, D. (2006). Treating preschool children who stutter: Description and Preliminary evaluation of a family-focused treatment approach. *Language, Speech, and Hearing Services in Schools, 37*, 118–136.

Zengin-Bolatkale, H., Conture, E., Key, A., Walden, T., & Jones, R. (2018). Cortical associates of emotional reactivity and regulation in childhood stuttering. *Journal of Fluency Disorders, 56*, 81–99.

Kerianne C. Druker is a PhD candidate in the School of Occupational Therapy, Social Work and Speech Pathology at Curtin University of Technology, Perth, Western Australia. She is a certified practising speech pathologist. Her clinical and research areas of interest include treatment of fluency disorders in paediatric populations and examining the role that self-regulation plays in early-onset stuttering.

Neville W. Hennessey is a lecturer in the School of Occupational Therapy, Social Work and Speech Pathology at Curtin University of Technology, Perth, Western Australia. His current research interests include stuttering, speech disorders in children and adults, language and literacy development and disorders, and psycho-linguistics.

Trevor G. Mazzucchelli is a registered clinical psychologist who has specialised in parenting interventions for families of children with behaviour disorders. He is also a senior lecturer in clinical psychology in the School of Psychology at Curtin University of Technology, Perth, Western Australia.

Janet M. Beilby is an Associate Professor of Stuttering Disorders in the School of Occupational Therapy, Social Work and Speech Pathology at Curtin University of Technology, Perth, Western Australia. Her current research interests include parent-administered early treatment initiatives, adolescent stuttering treatment programs and the psychosocial impact of stuttering disorders.

Link between Paper 1 and Paper 2

Paper 1 (presented in Chapter 3 of this thesis) identified 49.7% of CWS present to clinic with elevated ADHD symptoms. This highlights that half of CWS on a clinical caseload may present with co-occurring behaviour challenges, necessitating consideration of these symptoms in comprehensive and holistic management of this disorder.

In Paper 1, it was also determined that the presence of elevated ADHD symptoms had a significant negative impact on these children's responsiveness to, and success with, stuttering treatment. Despite the impact of these behaviour challenges on stuttering treatment outcomes, studies to date have not attempted to address these behavioural self-regulation challenges as a component of developmental stuttering management, for CWS particularly challenged with these issues. This finding provides support for the development and evaluation of an integrated stuttering and self-regulation program for this subgroup of CWS, to primarily determine if indirectly improving these children's behavioural self-regulation skills enhances their stuttering treatment outcomes. Paper 2 (presented in Chapter 4 of this thesis) reports on the implementation and evaluation of an integrated stuttering and self-regulation program for this subgroup of CWS, in a three-pronged controlled clinical trial.

Chapter 4

Paper 2: An evaluation of an integrated stuttering and parent-administered self-regulation program for early developmental stuttering disorders

Research Article

An Evaluation of an Integrated Stuttering and Parent-Administered Self-Regulation Program for Early Developmental Stuttering Disorders

Kerianne Druker,[a] Trevor Mazzucchelli,[b,c] iD
Neville Hennessey,[a] iD and Janet Beilby[a] iD

Purpose: This study reports findings from a clinical trial that implemented an early stuttering treatment program integrated with evidence-based parenting support (EBPS) to children who stutter (CWS) with concomitant self-regulation challenges manifested in elevated attention-deficit/hyperactivity disorder (eADHD) symptoms and compared those outcomes to CWS receiving stuttering treatment without EBPS.
Method: Participants were 76 preschool CWS and their parent(s). Thirty-six of these children presented with eADHD and were quasirandomized into two groups: stuttering treatment only (eADHD$_{standard}$) or stuttering treatment integrated with EBPS (eADHD$_{integrated}$). The remaining children did not meet criteria for eADHD symptoms and received stuttering treatment only (No-eADHD$_{standard}$). Pre, post, and 3-month follow-up measures of stuttering treatment outcomes as well as treatment effects on measures of child behavior difficulties and parenting practices were examined.

Results: Significant reduction in stuttering was found for all groups. However, the eADHD$_{integrated}$ group showed a greater reduction in stuttering frequency than the eADHD$_{standard}$ group, and at follow-up, stuttering frequencies in the eADHD$_{integrated}$ group matched those of children in the No-eADHD$_{standard}$ group, while stuttering in the eADHD$_{standard}$ group remained significantly higher. Children with eADHD symptoms who received the integrated program also required significantly less stuttering intervention time than those children with eADHD symptoms who received stuttering treatment only. Families in the eADHD$_{integrated}$ group reported large and significant improvements in child behavior and parenting practices.
Conclusion: This study provides support for an early treatment program for CWS. The integrated stuttering and self-regulation management program for CWS with eADHD symptoms proved successful for fluency and behavioral improvements, which were sustained at follow-up.

E arly intervention is important to facilitate positive treatment outcomes for preschool children who stutter (CWS; Oberklaid, 2014; Reilly et al., 2013). A recent study has demonstrated that one quarter of CWS did not respond to contemporary stuttering treatment, regardless of whether direct or indirect approaches were

implemented (de Sonneville-Koedoot et al., 2015). Furthermore, substantive treatment dropout rates have been reported in preschool stuttering interventions (McCulloch et al., 2017; Trajkovski et al., 2019). The implications of these findings are significant when paired with evidence that preschool-age children have the best chance of reducing or eliminating long-term stuttering with early intervention (McCulloch et al., 2017). Therefore, research indications

[a]School of Occupational Therapy, Social Work and Speech Pathology, Curtin University, Perth, Western Australia, Australia
[b]School of Psychology, Curtin University, Perth, Western Australia, Australia
[c]Parenting and Family Support Centre, School of Psychology, The University of Queensland, Brisbane, Australia

Correspondence to Janet Beilby: J.Beilby@curtin.edu.au

endorse the need for a more individually tailored approach to intervention to address concomitant factors in clinic profiles for a cohort of CWS whose engagement with and/or responsiveness to treatment is hindered (Donaghy & Smith, 2016).

Stuttering and Self-Regulation

One factor that has received significant attention in the early stuttering literature in terms of its possible impact on the manifestation and prognosis of the disorder is self-regulation (Eggers et al., 2010; Jones et al., 2014). Self-regulation skills are recognized as a component of temperament that develops over time and is influenced by caregiver interactions (Eisenberg & Sulik, 2012; Sameroff, 2010). The theoretical concept of self-regulation broadly refers to the capacity of a child to execute adaptive and goal-directed behaviors (Sameroff, 2010; Sanders & Mazzucchelli, 2018; Tominey & McClelland, 2011). Self-regulatory capacities include effortful control, which is the ability to inhibit prepotent behaviors, shift and focus attention as required, and activate goal-oriented actions when there is a propensity to avoid them (Eisenberg & Sulik, 2012; Posner & Rothbart, 2009). Although effortful control is often automatic, aligned with temperament, there is a component that is subject to conscious control (Eisenberg & Spinrad, 2004).

In the dual diathesis–stressor (DD-S) model of early stuttering, it is recognized that emotion diatheses and stressors have an impact on developmental stuttering (Walden et al., 2012). In particular, the emotion diathesis comprises an individual's proclivities for emotion reactivity and regulation (Walden et al., 2012). Emotional reactivity is characterized by a level of sensitivity to emotional stimuli resulting from involuntary or "bottom-up" processing (Sanchez et al., 2019). Emotional control or regulation depends on more voluntary or "top-down" processing that is responsible for inhibiting inappropriate responses when required (Sanchez et al., 2019). Emotional stressors refer to environmental changes that elicit emotion (Walden et al., 2012). High emotional reactivity and difficulty in emotional regulation can result in disruptive behaviors, such as impulsivity, avoidance or aggression when dealing with emotionally stimulating situations, and difficulty calming down when upset (Clark et al., 2002; Miller et al., 2004; Posner & Rothbart, 2009). The DD-S model, therefore, provides theoretical support for the notion that increased emotional reactivity and lower emotional regulation are linked to greater stuttering severity and frequency (Walden et al., 2012).

The demands and capacities model (DCM; Starkweather, 1987) also recognizes the possible significance of self-regulation in the manifestation of stuttering disorders. In this model, stuttering occurs as a result of the imbalance between cognitive, linguistic, motoric, and socio-emotional demands in the child and their ultimate capacity to manage these demands and produce resultant fluent speech (Starkweather, 1987; Starkweather et al., 1997). CWS with concomitant self-regulation deficits are likely to respond to environmental changes with increased emotion and

volatility (Eisenberg & Sulik, 2012). These volatile, emotional reactions place increased demands on the speech motor system of the CWS, subsequently reducing their capacity for controlled, fluent speech production (Smith & Weber, 2017; Starkweather, 2002).

CWS have been found to exhibit poorer self-regulation skills and increased emotional reactivity compared to their fluent peers (Eggers et al., 2010; Jones et al., 2014; Karrass et al., 2006). Such results align with the principles inherent within the DD-S model and the DCM. Such alignment is further supported by research showing that children with more severe stuttered speech severity have the most difficulty with self-regulation skills (Kraft et al., 2014, 2019).

The DCM proposes that improvement in the motoric, linguistic, socio-emotional, and/or cognitive function in a child who stutters, and/or through facilitation of the child's environment, results in a reduction or recovery from stuttering (Franken & Putker-de Bruijn, 2007). This assertion provides support for addressing self-regulation abilities in CWS with self-regulation deficits, to reduce the child's vulnerability to fluency destabilization that arises as a result of exceeding their capacity to manage socio-emotional demands.

Stuttering and Attention-Deficit/Hyperactivity Disorder

According to the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (American Psychiatric Association, 2013), attention-deficit/hyperactivity disorder (ADHD) is conceptualized as a disorder of self-regulation (Barkley, 2013; Huguet et al., 2019; Reid et al., 2005; Spencer et al., 2011). Individuals with ADHD display emotional reactivity traits and exhibit difficulties with emotional control or regulation (Petrovic & Castellanos, 2016). Therefore, the challenges in self-regulation for CWS appear to overlap, to some degree, with the behavioral symptoms demonstrated in ADHD given that a primary challenge in ADHD is that of behavioral dysregulation (Barkley, 2013).

Although there is no known evidence to suggest that CWS are at a greater risk of exhibiting clinical ADHD diagnoses, studies have found that CWS do experience greater effortful control difficulties (Ntourou et al., 2018) and elevated or subclinical ADHD symptoms compared to their fluent peers (Alm & Risberg, 2007; Donaher & Richels, 2012; Felsenfeld et al., 2010). Subclinical symptoms refer to a child exhibiting ADHD symptoms that do not necessarily meet criteria for clinical diagnosis, but still have an impact on the child's functioning in daily activities (Donaher & Richels, 2012).

In a recent large-scale study (Druker, Hennessey, et al., 2019), it was found that 50% of CWS who presented to a stuttering treatment clinic, in addition to their stuttered speech diagnosis, displayed concomitant elevated ADHD (eADHD) symptoms. In addition, retrospective analysis found that these CWS required 25% more clinical intervention time to achieve successful fluency outcomes, compared to CWS without eADHD symptoms (Druker, Hennessey,

et al., 2019). These findings are supportive of genotypic research with twins that has suggested a possible subgroup or cohort of CWS to exist with concomitant ADHD symptoms (Felsenfeld et al., 2010), as well as research highlighting the effect of "attending disorders" on the prognosis for stuttered speech outcomes (Riley & Riley, 2000).

Addressing ADHD Symptoms in CWS

Interventions that have been shown to be effective in promoting self-regulation skills for children with disruptive behaviors (Norris, 2016), including children with ADHD (Barkley, 2013; Sanders, Kirby, et al., 2014; Webster-Stratton et al., 2013), are also likely to be effective for those presenting with subclinical ADHD symptoms. While selected approaches to the management of childhood stuttering recognize the importance of addressing child self-regulation through provision of parent support (Millard & Davis, 2016; Starkweather et al., 1997), to the authors' knowledge, no previous intervention study has attempted to evaluate the effectiveness of an integrated self-regulation and stuttering treatment program for CWS specifically challenged with self-regulation difficulties or eADHD symptoms.

Evidence-based parenting support (EBPS) has been shown to be effective and beneficial for a number of child health and well-being concerns (e.g., Bjornebekk et al., 2015; Bradshaw et al., 2019; Grey et al., 2011; Sanders & Mazzucchelli, 2018), including the reduction in disruptive childhood behaviors (Sanders & Mazzucchelli, 2018). Although EBPS has not yet been trialed with parents and families of CWS with eADHD symptoms, based on research to date, it is anticipated that it may yield positive outcomes for this population through implementation of parenting strategies such as positive reinforcement for compliance as well as consistent, appropriate behavioral consequences (Bor et al., 2002). In addition, improvements in parent–child interactions are a key focus of EBPS, which is anticipated to increase the child's cooperation to act on and be guided by parent-suggested strategies to facilitate fluent speech production (Bor et al., 2002; Sanders, Kirby, et al., 2014).

This Study

Early successful management of developmental stuttering is desirable and important for CWS and their families (Beilby, 2014; Langevin et al., 2010). In addition, poorer self-regulation skills in childhood, manifested in eADHD symptoms, are prevalent in childhood stuttering (Druker, Hennessey, et al., 2019). These symptoms are hypothesized to play a key role in sustaining fluency difficulties, given evidence that they negatively impact stuttering treatment responsiveness and long-term fluency outcomes (Druker, Hennessey, et al., 2019; Riley & Riley, 2000).

The goal of strengthening neural networks for speech motor planning and execution, which are robust to demands of fluctuating emotional state, has been highlighted in contemporary multifactorial models of early stuttering (Smith & Weber, 2016, 2017). Furthermore, improving self-regulation at preschool age may moderate possible negative long-term emotional, social, and occupational outcomes that have been demonstrated in children with behavioral self-regulation challenges (Moffitt et al., 2011). Therefore, the primary aim of the study was to (a) evaluate the effectiveness of an integrated self-regulation and stuttering treatment program, for a cohort of CWS with eADHD symptoms, on stuttering treatment outcomes (i.e., stuttered speech severity and treatment responsiveness). Secondary aims of the study were to (b) evaluate the impact of this integrated program on parent and child psychosocial outcomes and (c) compare the presentation characteristics (including pretreatment severity of stuttered speech and psychosocial measures) and treatment outcomes of CWS with and without concomitant eADHD symptoms. Given the Curtin University Stuttering Program (CUSP) was the stuttering intervention administered to all participants in the current study, a final secondary aim of this article was to (d) evaluate the effectiveness of the CUSP for the treatment of early developmental stuttering disorders.

The intervention group comprised CWS who met criteria for eADHD symptoms (Donaher & Richels, 2012; Druker, Hennessey, et al., 2019) and received an integrated parent-administered self-regulation and standard stuttering treatment program (eADHD$_{integrated}$). The primary comparison group comprised CWS who also met criteria for eADHD but received standard stuttering treatment only (eADHD$_{standard}$). To address the study's third aim, the eADHD children were compared to a third group of CWS who did not meet criteria for eADHD and who received the standard stuttering treatment (No-eADHD$_{standard}$). Standard stuttering treatment for all three groups refers to the CUSP, which was implemented consistently across all speech pathology clinics from which participants were recruited. The intervention component used to address self-regulation difficulties in CWS with eADHD was the Triple P—Positive Parenting Program (Sanders & Mazzucchelli, 2018).

Supportive of the primary aim of the study, it was hypothesized that reducing the impact of disruptive behaviors, and facilitating self-regulation, would increase the capacity of CWS with eADHD to achieve and maintain speech fluency. The prediction was that improved stuttering treatment outcomes would result for the eADHD$_{integrated}$ group in terms of greater reduction in percentage of syllables stuttered (%SS) from pretreatment to posttreatment and 3-month follow-up. We also tested whether parental participation in the Triple P will reduce the number of face-to-face stuttering treatment sessions required to reach discharge for the CWS in the eADHD$_{integrated}$ group when compared to the eADHD$_{standard}$ group. This will provide evidence, at least for children with eADHD symptoms, that an additional EBPS program designed to help parents facilitate their child's behavioral self-regulation can improve responsiveness to a standard stuttering treatment that is focused primarily on managing the child's stutter.

Supportive of the study's secondary aim of evaluating the impact of the integrated program on psychosocial outcomes, it was predicted that, given the documented effectiveness of the Triple P in successfully reducing childhood disruptive behaviors, there would be a reduction at posttreatment and 3-month follow-up in ADHD symptoms, as measured by the ADHD Rating Scale-IV: Home Version (McGoey et al., 2007; Power et al., 2001), as well as a reduction in social–emotional challenges, as measured by the Strengths and Difficulties Questionnaire (SDQ; R. Goodman, 1997) for the eADHD$_{integrated}$ group compared to the eADHD$_{standard}$ group. In addition, it was anticipated that parents of the children in the eADHD$_{integrated}$ group, compared to those in the eADHD$_{standard}$ group, would show improved parenting skills at posttreatment and follow-up as measured by the Parenting and Family Adjustment Scales (PAFAS; Sanders, Morawska, et al., 2014).

Finally, even though the retrospective study of Druker, Hennessey, et al. (2019) did not find that CWS with eADHD presented with more severe stuttering pretreatment than children without eADHD, such a potential difference is consistent with current theoretical models of developmental stuttering, which propose that the capacity of CWS to achieve fluent speech may be compromised by stressors including deficits in self-regulation (Walden et al., 2012). Therefore, in line with the secondary aim of comparing presentation characteristics between groups, it was predicted that children in this study in the No-eADHD group would show less severe stuttered speech at pretreatment than both eADHD groups. In addition, better treatment outcomes—that is, lower %SS and fewer treatment sessions—were anticipated for the No-eADHD group, at least with respect to the eADHD$_{standard}$ group who did not receive the integrated intervention.

To provide additional support for the assertion that CWS with eADHD have difficulties in self-regulation, the No-eADHD$_{standard}$ group were expected to score higher on the effortful control subscale of the Child Behavior Questionnaire (CBQ-EC; Rothbart et al., 2001) when compared to both eADHD groups. The effortful control subscale assesses aspects of self-regulation including inhibitory control and capacity to focus attention (Rothbart et al., 2001) and has been shown to be associated with increased severity of childhood stuttering (Kraft et al., 2014, 2019). Because effortful control is viewed as a component of child temperament (Rothbart et al., 2011) and may, therefore, be relatively stable over time, it was anticipated that the beneficial intervention effects on this particular measure might be limited.

Finally, in line with the study's final secondary aim of evaluating the effectiveness of the CUSP, it was predicted that the CUSP would be effective in significantly reducing stuttered speech severity from pre- to posttreatment, with outcomes maintained at follow-up (Druker, Hennessey, et al., 2019; Druker, Mazzucchelli, Hennessey, & Beilby, 2019).

In summary, evidence-based, established stuttering treatments have not proven successful for all CWS, and high dropout attrition rates have been reported (de Sonneville-Koedoot et al., 2015; McCulloch et al., 2017; Trajkovski et al., 2019). The present research builds on a previous retrospective study (Druker, Hennessey, et al., 2019), which demonstrated how difficulty in self-regulation for CWS is a factor that negatively affects standard stuttering treatment engagement and responsiveness. In the current study, a prospective clinical trial was implemented to investigate whether self-regulation difficulties (as assessed by the symptomology of ADHD) are associated with poorer treatment outcomes when compared to those for preschool CWS without that symptomology. In addition, this study evaluated if stuttering treatment outcomes along with eADHD symptomatology can be significantly improved by combining a standard stuttering treatment with an EBPS program designed specifically to address poor self-regulation skills in CWS.

Method

Participants

Participants comprised 77 preschool children aged from 2;0 to 5;3 (years;months) with clinically diagnosed stuttered speech disorders and their parent(s). Seventy-three CWS and their families lived in the Perth metropolitan area, and three resided in rural Western Australia (> 80 km from the city center). One international family who lived in Ireland was included, with all treatments delivered via telehealth.

The following inclusion criteria were applied: (a) CWS having hearing within normal limits, based on parent report; (b) presence of developmental stuttering disorder, as diagnosed by a fluency specialist speech pathologist; and (c) English as the primary language spoken in the family. Exclusion criteria included (a) family already receiving regular intervention for child behavioral problems (including pharmacological) and/or parenting support and (b) child having a clinical diagnosis of autism spectrum disorder at the time of initial testing.

Between-groups analysis (ANOVA and chi-square tests) showed no significant differences ($ps > .23$) between the treatment groups in age at stuttering onset, weeks between stuttering onset and commencement of treatment, chronological age, sex, family history of developmental stuttering disorder, or phonological and language abilities (see Table 1). Engagement with and commitment to the intervention is evidenced through the low attrition rates of 1.32% at posttreatment and 5.26% at follow-up.

Primary Outcome Measures

%SS was used as an outcome measure of severity of stuttered speech behaviors, consistent with previous behavioral stuttering treatment research (Karimi et al., 2014). Pretreatment, posttreatment, and follow-up %SS scores were measured by calculating the %SS, out of a total of 2,000 syllables produced, using the Fluency Rater Application (ForFluency, 2012). Severity ratings were calculated based on within-clinic samples obtained through naturalistic

Table 1. Participant demographics.

Variable	No-eADHD$_{standard}$ $n = 40$	eADHD$_{standard}$ $n = 17$	eADHD$_{integrated}$ $n = 19$
Age at onset, M (SD)	2.91 (0.82)	3.33 (0.94)	2.97 (0.76)
Weeks since onset, M (SD)	34.71 (44.30)	43.88 (48.99)	31.25 (33.23)
Chronological age in years, M (SD)	3.56 (2.33)	4.18 (1.02)	3.57 (.90)
PCC, M (SD)	45.18 (30.36)	35.64 (28.25)	42.90 (27.84)
CLS, M (SD)	66.28 (23.75)	63.86 (23.40)	58.68 (29.40)
Sex (% male)	68.3	87.5	75.0
Family history (% positive)	85.4	68.8	85.0

Note. PCC score refers to percentile rankings calculated from the percent consonants correct measure extracted from the Phonology subtest of the Diagnostic Evaluation of Articulation and Phonology (Dodd et al., 2002). CLS score refers to percentile rankings calculated from the Core Language Score of the Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Wiig et al., 2006). No-eADHD$_{standard}$ = children who stutter without elevated attention-deficit/hyperactivity disorder (ADHD) symptoms who received stuttering treatment only; eADHD$_{standard}$ = children who stutter with elevated ADHD symptoms who received stuttering treatment only; eADHD$_{integrated}$ = children who stutter with elevated ADHD symptoms who received the integrated self-regulation and stuttering treatment program; PCC = percent consonants correct; CLS = Core Language Score.

conversation with the treating clinician using a wordless picture book and toy animals. Stuttered speech behaviors included part-word repetitions, sound prolongations, and audible or inaudible blocks (Guitar, 2014; Smith & Weber, 2017). Reliability checks for %SS ratings were conducted on 10% of the total sample by the fourth author. Pearson's product–moment correlation analysis identified a high level of inter-rater reliability, $r = .94$, $p < .01$.

Responsiveness to stuttering treatment was measured by the number of stuttering treatment sessions required for the child to reach discharge criteria. Discharge criteria in the current study included a consistent stuttered speech severity rating of 2%–3% stuttered syllables or less across a variety of within and beyond clinic speech situations and when parents felt satisfied with their child's level of fluency and their ability to manage any remaining minor disfluencies on their own (Millard et al., 2018). If there were any concerns about their children's fluency after discharge, all parents were encouraged to recontact the clinic from which they were recruited from (Millard et al., 2018).

Secondary Outcome Measures

The ADHD Rating Scale-IV: Home Version (McGoey et al., 2007; Power et al., 2001) is an 18-item questionnaire extracted from the ADHD symptoms list specified in the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* diagnostic criteria, used for the screening of ADHD (American Psychiatric Association, 2013). The scale comprises nine items measuring inattention and nine items measuring hyperactivity/impulsivity (DuPaul et al., 1998). Parents rate each item on a 4-point scale, ranging from *never or rarely* (0) to *very often* (3). The scale has strong concurrent validity and internal consistency, with correlations ranging from .8 to .87 (DuPaul et al., 1998; McGoey et al., 2007). Reliability coefficients ranged from .78 to .95, indicating strong test–retest reliability (DuPaul et al., 1998; McGoey et al., 2007). Given the age range of participants in the

study, two versions of the scale were administered: one version for children aged 2;0–4;11 (McGoey et al., 2007) and the second version for children aged 5;0–6;11 (Power et al., 2001).

The scale was used to initially identify the presence of eADHD, prior to allocating those CWS into the integrated and standard treatment groups (see Figure 1) following the procedure detailed below. Cutoff criteria applied for groups were based on Donaher and Richels (2012) and Druker, Hennessey, et al. (2019): Children were classified as presenting with eADHD if they scored a rating of 2 (*often*) or greater on at least two inattention items or hyperactivity/impulsivity items as well as an overall scale score within the top 20th percentile for the child's age and sex (DuPaul et al., 1998; McGoey et al., 2007).

The SDQ (R. Goodman, 1997) is a 25-item parent report instrument measuring parent perceptions of child behavioral and emotional problems. It has been used to measure socio-emotional development in CWS (McAllister, 2016), screen for both ADHD (Ullebø et al., 2011) and childhood mental health problems (R. Goodman, Renfrew, & Mullick, 2000), and measure change in child behavior following parenting interventions (Fujiwara et al., 2011). To reflect the presence of overall behavioral or psychological problems (R. Goodman, 1997), the total difficulties score was obtained based on adding together raw scores from the emotional problems, conduct problems, hyperactivity, and peer problems SDQ subscales. Each item was rated on a 3-point scale: *not true* (0), *somewhat true* (1), and *certainly true* (2). The mean Cronbach's α across subscales was found to be greater than .7, indicating satisfactory internal consistency (Goodman, 1997). Test–retest coefficients ranged from .57 to .82 across a 4- to 6-month interval, indicating satisfactory reliability (R. Goodman, Ford, et al., 2000).

Two versions of the scale were administered depending on participant age: one version for children up to 5 years of age and one version for 6-year-old participants. Items are identical across both versions, with the exception of

Figure 1. CONSORT flow diagram. ADHD = attention-deficit/hyperactivity disorder; CWS = children who stutter; eADHD = elevated attention-deficit/hyperactivity disorder; eADHD_integrated = children who stutter with elevated ADHD symptoms who received the integrated self-regulation and stuttering treatment program; eADHD_standard = children who stutter with elevated ADHD symptoms who received stuttering treatment only; EBPS = evidence-based parenting support; No-eADHD_standard = children who stutter without elevated ADHD symptoms who received stuttering treatment only; %SS = percentage of syllables stuttered.
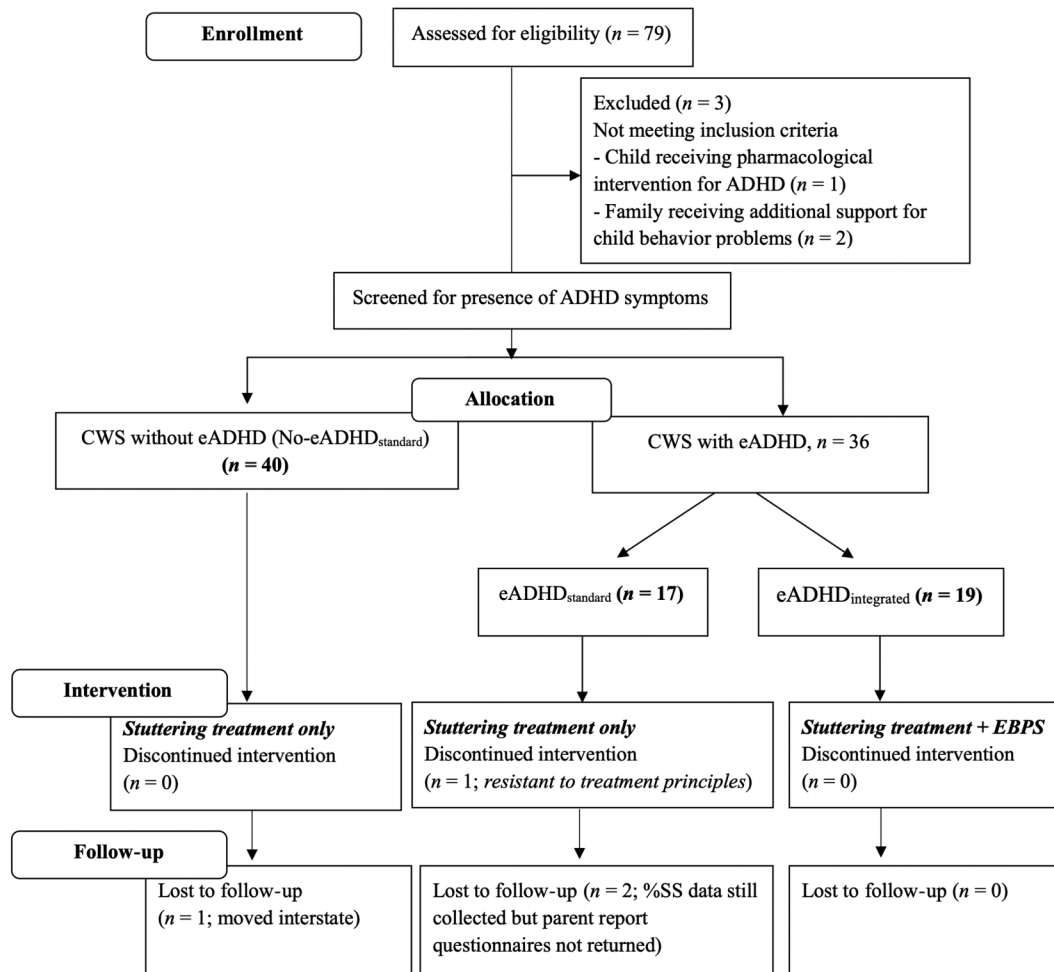
minor moderated wording of two items on the conduct scale and one item on the hyperactivity scale, for the younger age group (R. Goodman, 1997).

The CBQ-EC (Rothbart et al., 2001) is a parent report measure of child temperament associated with self-regulation. In particular, the capacity of the child to focus attention and withhold or inhibit dominant responses is evaluated (Posner & Rothbart, 2009). The CBQ-EC comprises four subscales (attention focusing, inhibitory control, low intensity pleasure, and perceptual sensitivity) and consists of 47 items rated on a 7-point Likert scale from *extremely untrue* (1) to *extremely true* (7). Cronbach's α for all subscales of interest was > .7, indicating satisfactory internal consistency (Rothbart et al., 2001). The mean test–retest reliability coefficient was acceptable at .64 (Rothbart et al., 2001).

The PAFAS (Sanders, Morawska, et al., 2014) is a 30-item parent report inventory assessing parenting practices, parental adjustment, and family relationships—domains that have been found to be predictive of mental health outcomes in children (Shaw et al., 2001). Items are scored on a 4-point Likert scale ranging from *not true of me at all* (0) to *true of me very much, or most of the time* (3). The scale includes two overarching domains: parenting practices (comprising parental consistency, coercive parenting, positive encouragement, and parent–child relationship subscales) and family adjustment (comprising parental adjustment, family relationships, and parental teamwork subscales). A total score for each domain was calculated by adding together raw scores from each of the comprising subscales. Lower scores indicate more desirable parenting practices and family adjustment. This measure has adequate predictive and construct validity, strong internal consistency (> .70 for all subscales), and good test–retest reliability (> .60; Sanders, Morawska, et al., 2014).

Procedure

Ethics approval was obtained for this study through the Curtin University Human Research Ethics Committee. Participants were recruited from the waitlists of the Curtin University Stuttering Treatment Clinic (CSTC) and an affiliated off-site private specialist fluency clinic and were enrolled between October 2017 and December 2018. Eighty participant dyads (CWS and their parent/s) were assessed for eligibility. Seventy-seven met inclusion criteria, and the children were then further screened for the presence of eADHD symptoms, assessed by parental responses on the ADHD Rating Scale (Druker, Hennessey, et al., 2019; see Figure 1).

Thirty-six children (47%) met criteria for eADHD symptoms and were allocated into either the eADHD$_{standard}$ ($n = 16$) or eADHD$_{integrated}$ ($n = 20$) group using a random draw method with the following exceptions. Five parents had specifically requested allocation to the eADHD$_{integrated}$ group after reading the information sheet regarding this study in the clinic waiting room prior to treatment commencing and researching the Triple P website. Two parents who were initially randomly allocated to the eADHD$_{integrated}$ group did not feel the additional parenting support was warranted and were reallocated to the eADHD$_{standard}$ group. Although randomization to group was planned for all participants, consideration was given to real-world, ecologically valid factors when allowing this small element of self-selection to occur.

The speech and language skills of each child participant were assessed within clinic or during a home visit between Fluency Sessions 1 and 3, by the first author (including telehealth administration for the single international child). To assess phonological development, percent consonants correct was calculated using the Phonology subtest of the Diagnostic Evaluation of Articulation and Phonology and converted to a percentile ranking (Dodd et al., 2002). To assess overall language development, a core language score was calculated using the Clinical Evaluation of Language Fundamentals Preschool–Second Edition and converted to a percentile ranking (Wiig et al., 2006). The total duration of the speech and language screening assessment lasted approximately 30 min.

All participants across the three groups received the CUSP early stuttering intervention, with treating clinicians blinded to the participants' group allocation. In addition to the CUSP stuttering treatment, parents in the eADHD$_{integrated}$ group received an EBPS program (Sanders, Kirby, et al., 2014) adapted to the needs of the stuttered speech disorder. See Figure 1 for a diagrammatic representation of the treatment implemented in each intervention group.

The EBPS commenced between Fluency Sessions 1 and 3, depending on parent availability. To assess treatment effects and maintenance of treatment outcomes, %SS at pretreatment, post-treatment, and follow up were collected from all three groups, as well as the number of stuttering treatment sessions. Additional psychosocial measures, including the ADHD Rating Scale-IV: Home Version, SDQ, CBQ-EC, and PAFAS, were collected at similar time points.

Interventions

CUSP

All children received the CUSP, at the CSTC, via telehealth or in the private affiliated off-site clinic (Druker, Hennessey, et al., 2019; Druker, Mazzucchelli, Hennessey, & Beilby, 2019). The clinics followed the same procedures and implemented the same stuttering therapy approach, which is an amalgamation of a direct behavioral component in conjunction with indirect principles and strategies. For example, parents are trained to modify linguistic, cognitive, and motoric environmental demands to support fluency and use fluency facilitation techniques as appropriate for the type of stuttered speech behaviors the child is demonstrating and the conversation contexts in which they are communicating (Millard et al., 2008; Starkweather, 1987). Parents in all three groups were trained to implement these combined techniques online throughout their daily conversations with their child (see Table 2 for stuttering management strategies implemented based on stuttering typography).[1] Across all three groups, the average length of stuttering treatment was 11 sessions ($SD = 4.92$, range: 3–30).

EBPS

EBPS based on social learning principles has been trialed comprehensively and has yielded positive parenting and behavioral outcomes in children with behavior problems (Sanders, Kirby, et al., 2014). The Triple P is an EBPS program designed to support parents and caregivers to prevent and manage behavioral and emotional difficulties in children aged 2–12 years (Sanders & Mazzucchelli, 2018). The program promotes positive and self-regulated parenting and creates supportive environments to develop children's self-regulation skills and competencies (Sanders & Mazzucchelli, 2018). Additional aims facilitated by this program include reduction of parenting stress, improvement in communication between parents, and increased parental self-efficacy (Sanders & Mazzucchelli, 2018).

Examples of positive parenting strategies include spending one-on-one time with the child, providing praise for desirable behavior, giving positive attention to the child, incidental teaching, use of directed discussion for rule breaking, planned ignoring for minor behavior problems, and provision of logical consequences for undesirable behavior (Markie-Dadds et al., 2015).

Given its strong supporting evidence base (Sanders, Kirby, et al., 2014; Sanders & Mazzucchelli, 2018), the Triple P was selected as the EBPS component of this study's integrated program. An integrated set of suggestions, in the form of a special "tip sheet," was created that incorporated key Triple P principles with strategies overlaid for fluency

---

[1]Additional inquiries about implementation of the CUSP can be directed to the corresponding author.

Table 2. Stuttering management strategies based on stuttering typography.

	Typography		
Strategies	Repetitions	Prolongations	Blocks
Labeling	That was a bit bumpy	That was a bit too long	That sounded a bit stuck
Suggestions	Try saying it just once	Try using shorter sounds	Try just saying…
Neutral interjections	Oops, careful	Oops, careful	Oops, careful
Neutral questions	Pardon, what's that?	Pardon, what's that?	Pardon, what's that?
Helping out techniques			Say the word with the child. Use rising intonation and inflection.
			Provide choices. Simplify the exchange.
			Slow down parent rate of speech.
			Aim for a shift in the nature of the stutter down from blocks to easier versions of the stutter such as whole-word repetitions.

management, uniquely developed by the authors of the current study (Druker et al., 2018). This tip sheet is of similar nature to tip sheets used in previous studies involving adaption of the Triple P to specific populations (e.g., Hoath & Sanders, 2002).

In the current study, the Triple P component comprised eight sessions delivered individually and extending over an average of 10 weeks. Sessions were delivered by the first author, a trained and accredited Level 4 Group Triple P practitioner. Group Triple P principles were adapted to be delivered individually and predominantly via Skype, in response to parents' feedback regarding access to the program. This flexible adaption is consistent with the content-adherent, yet process-flexible, approach advocated by the program developers (Mazzucchelli & Sanders, 2010).

An initial 45-min face-to-face introductory session highlighted links between self-regulation and stuttering and how positive parenting practices were relevant to addressing both. The tip sheet was used to guide the discussion in this session. Four weekly 60-min Skype sessions followed and provided parents with information on positive parenting practices, using a standardized workbook to guide parent discussions (Markie-Dadds et al., 2015). Three 20-min weekly telephone consultations were then provided to support parents in implementing parenting strategies (Markie-Dadds et al., 2015).

Given parents' original concern related to their children's stuttering, examples of how EBPS principles could be applied to stuttering were provided to enable parents to perceive the EBPS component as relevant and acceptable for them and their children. It may be noted, however, that the primary focus of the EBPS sessions was on positive parenting practices to facilitate child self-regulation across a range of behaviors of concern, not just stuttering. However, while this does suggest the parents of the children in the eADHD$_{integrated}$ group potentially received a greater dosage of clinical advice regarding their children's stutter, compared to parents of the eADHD$_{standard}$ children, we did not adjust the primary outcome measure of treatment responsiveness, the total number of stuttering treatment sessions, to account for this input. It was not possible to identify how much time from the eight EBPS sessions was focused on stuttering to

make an adjustment, and treating all eight EBPS sessions as stuttering treatment sessions is inappropriate as a measure of stuttering treatment responsiveness because self-regulation of stuttering behaviors was only a component of the entire EBPS program. We consider this point in more detail in the Discussion section. Nevertheless, we do report an analysis with both stuttering treatment and EBPS sessions combined as a comparison of total time commitment associated with the different treatment conditions in the study.

Treatment fidelity. To ensure fidelity of the CUSP, each speech pathologist completed standard checklists of each session, including type and frequency of stuttering treatment principles implemented. All sessions were video-recorded, and 10% of stuttering treatment sessions were randomly selected and independently rated by an external rater. Interrater reliability was calculated at 85%. To ensure fidelity of the Triple P component of the integrated intervention, the facilitator rated all Triple P sessions, and on average, 84% of the intended session content was covered. Ten percent of these sessions were randomly selected and rated by an independent rater with an interrater agreement of 91%.

Data Analysis

Separate generalized linear mixed models (GLMMs), implemented through SPSS Statistics (Version 25, IBM Corp. 2017) GENLINMIXED procedure, were used to test for group differences and time-related changes in each outcome measure. GLMM is a robust analysis that has the advantage of accommodating a variety of response variables including nonnormal distributions. The models used to investigate the effect of the integrated treatment for the CWS with eADHD symptomology included participants as a random effect, as well as the fixed effects of group (eADHD$_{integrated}$ vs. eADHD$_{standard}$) and time (e.g., pre- vs. posttreatment), and the Group × Time interaction. A significant interaction indicates the change from pretreatment to posttreatment, or from pretreatment to follow-up, varies across the two groups. Each interaction was interpreted following simple effects analysis using least significant

difference (LSD) contrasts of marginal means. Planned comparisons at each time point between the groups with and without eADHD were also undertaken using Bonferroni-corrected (two-tailed) LSD contrasts from GLMMs with participants as a random effect and group as a fixed effect. For some analyses, covariates were identified and therefore also included as fixed effects in the GLMM (see below). The effect size reported is partial eta squared (obtained from SPSS GLM analysis of variance), and the α level used throughout is .05.

Results

The Effect of the Integrated Treatment on Fluency Outcomes for CWS With eADHD

The group means of each outcome measure at pretreatment, posttreatment, and follow-up are reported in Table 3. Analyses of pretreatment group differences are reported first. LSD contrasts (two-tailed) showed that, when compared to the eADHD$_{standard}$ group, the eADHD$_{integrated}$ group had significantly higher ADHD-IV combined scores, $t(74) = 2.72$, $p = .008$; ADHD-IV inattentive scores, $t(74) = 2.90$, $p = .005$; and SDQ total scores, $t(74) = 4.48$, $p < .001$, as well as significantly higher PAFAS parenting practices total score, $t(74) = 3.27$, $p = .002$. There were no differences between the eADHD$_{integrated}$ and eADHD$_{standard}$ groups in baseline %SS, $t(74) = 1.79$, $p = .077$; ADHD hyperactive/impulsive scores, $t(74) = 1.15$, $p = .253$; Child Behavior Questionnaire CBQ total scores, $t(74) = 0.38$, $p = .704$; or PAFAS family adjustment scores, $t(74) = 1.47$, $p = .147$.

GLMM analysis was then used to test the study's primary hypothesis that the integrated program would enhance stuttering treatment outcomes for CWS with eADHD symptoms. None of the variables showing group differences between the eADHD$_{integrated}$ and eADHD$_{standard}$ groups pretreatment (ADHD-IV combined, ADHD-IV inattentive, SDQ total, PAFAS parenting practices) were significant predictors of %SS ($ps > .05$). Therefore, no covariates were included in the analysis with %SS as the outcome measure. The GLMM for %SS showed no overall group effect, $F(1, 67) = 1.94$, $p = .169$, $\eta^2 = .06$, but there was a significant effect of time showing a reduction in %SS posttreatment, collapsed across both groups, $F(1, 67) = 197.80$, $p < .001$, $\eta^2 = .85$. With regard to the study's secondary aim to evaluate the effectiveness of the CUSP, this large effect size supports the hypothesis that the CUSP is effective in reducing stuttered speech severity.

Importantly, a significant interaction between group and time was present, $F(1, 67) = 4.82$, $p = .032$, $\eta^2 = .13$. The interaction indicates the decrease in %SS from pre to post for the eADHD$_{integrated}$ group was significantly greater than that for the eADHD$_{standard}$ group (see Figure 2). It is notable that, while the %SS mean was numerically higher pretreatment for the eADHD$_{integrated}$ group, the eADHD$_{integrated}$ mean was lower posttreatment, although this difference was not statistically significant, $t(67) = 1.81$, $p = .074$. The results were similar for %SS when comparing

pretreatment with follow-up (see Figure 2). There was no main effect of group, $F < 1$, but there was a significant effect of time, $F(67) = 195.75$, $p < .001$, $\eta^2 = .63$, indicating the reduction in %SS for both groups combined was maintained, as well as a Group × Time interaction, $F(1, 67) = 5.66$, $p = .020$, $\eta^2 = .45$. The data, therefore, show a significantly greater change from pretreatment to follow-up for the eADHD$_{integrated}$ group. In support of this result, the eADHD$_{integrated}$ group was significantly lower in %SS at follow-up compared to the eADHD$_{standard}$ group, $t(67) = 2.44$, $p = .018$.

The number (and percentage) of CWS with eADHD who did not meet discharge criterion of 2–3 %SS at follow-up was calculated. Four of 16 participants in the eADHD$_{standard}$ group did not meet this criterion (25%), while only one of 20 did not meet discharge criterion in the eADHD$_{integrated}$ group (5%).

All three groups were combined in the GLMM evaluating number of stuttering treatment sessions. Because the No-eADHD group had significantly lower %SS pretreatment than the eADHD groups combined (see No-eADHD Group Outcomes and Comparisons With eADHD Groups [Secondary Aims c and d] section), and %SS was significantly associated with the number of treatment sessions prior to entering the group fixed effect, $F(1, 73) = 12.97$, $p = .001$, %SS was included as a covariate in the model. The covariate remained significant, $F(1, 71) = 12.66$, $p = .001$, and the group fixed effect was also statistically significant, $F(2, 71) = 3.84$, $p = .026$. Bonferroni-corrected LSD contrasts of estimated marginal means showed, after controlling for %SS, the eADHD$_{integrated}$ group (adjusted $M = 10.0$, $SEM = 0.7$) required significantly fewer stuttering treatment sessions to discharge compared to the eADHD$_{standard}$ group (adjusted $M = 14.0$, $SEM = 1.3$), $t(71) = 2.49$, $p = .030$.[2] The total time commitment, however, was significantly greater for the intervention group, compared to the standard stuttering treatment group, when the EBPS program sessions were combined with the number of stuttering sessions (adjusted $M = 17.9$, $SEM = 0.7$), $t(71) = 2.45$, $p = .017$. Comparisons with the No-eADHD$_{standard}$ group are reported in the No-eADHD Group Outcomes and Comparisons With eADHD Groups (Secondary Aims c and d) section.

Psychosocial Outcomes for eADHD Groups

Results reported in this section align with the study's Secondary Aim b.

ADHD Rating Scale-IV

The GLMM comparing the eADHD$_{integrated}$ and eADHD$_{standard}$ groups in their pre- and posttreatment ADHD-IV combined scores showed no significant effect of group, $F < 1$, but a significant effect of time, $F(1, 67) = 43.46$, $p < .001$, $\eta^2 = .55$, as well as an interaction between

---

[2]The following are the unadjusted mean number of stuttering treatment sessions for both groups: eADHD$_{standard}$, $M = 14.2$ ($SD = 6.1$); eADHD$_{integrated}$, $M = 11.0$ ($SD = 3.2$).

Table 3. Pre, post, and follow-up treatment means and standard deviations for each treatment group.

Outcome measure	Pretreatment[a] M (SD)	Posttreatment[b] M (SD)	Follow-up[c] M (SD)
Primary outcome			
%SS			
No-eADHD$_{standard}$	11.96 (7.32)	1.73 (.49)	1.48 (.55)
eADHD$_{standard}$	15.76 (7.81)	2.61 (1.52)	2.90 (1.76)
eADHD$_{integrated}$	19.92 (6.12)	1.91 (.64)	1.80 (0.77)
Secondary outcomes			
ADHD combined			
No-eADHD$_{standard}$	8.89 (4.68)	8.96 (5.30)	8.51 (5.48)
eADHD$_{standard}$	24.69 (3.98)	23.00 (9.13)	20.79 (6.54)
eADHD$_{integrated}$	30.15 (8.07)	15.32 (7.38)	14.58 (6.87)
ADHD inattentive			
No-eADHD$_{standard}$	3.93 (2.44)	4.44 (3.33)	4.10 (3.01)
eADHD$_{standard}$	10.81 (3.49)	10.75 (5.30)	9.64 (5.26)
eADHD$_{integrated}$	14.50 (4.37)	6.74 (4.43)	6.79 (3.78)
ADHD hyperactive/impulsive			
No-eADHD$_{standard}$	4.99 (2.98)	4.55 (3.13)	4.67 (3.41)
eADHD$_{standard}$	13.88 (3.32)	12.25 (5.26)	11.07 (3.27)
eADHD$_{integrated}$	15.55 (9.58)	8.58 (4.94)	7.53 (4.81)
SDQ total			
No-eADHD$_{standard}$	6.76 (3.63)	7.29 (4.19)	6.79 (4.62)
eADHD$_{standard}$	12.63 (3.85)	11.88 (4.43)	11.07 (3.58)
eADHD$_{integrated}$	19.90 (6.10)	10.05 (4.65)	10.05 (5.36)
CBQ-EC total			
No-eADHD$_{standard}$	5.25 (0.54)	5.46 (0.54)	5.57 (0.51)
eADHD$_{standard}$	4.68 (0.56)	4.77 (0.67)	4.88 (0.69)
eADHD$_{integrated}$	4.6 (0.51)	5.05 (0.48)	5.07 (0.56)
PAFAS parenting practices			
No-eADHD$_{standard}$	10.71 (4.68)	10.25 (5.26)	10.26 (4.15)
eADHD$_{standard}$	10.81 (4.78)	11.00 (3.29)	11.57 (3.78)
eADHD$_{integrated}$	16.15 (5.29)	6.58 (4.35)	6.74 (4.15)
PAFAS home adjustment			
No-eADHD$_{standard}$	5.34 (3.45)	5.78 (4.82)	5.54 (3.17)
eADHD$_{standard}$	9.19 (5.46)	12.00 (4.12)	10.71 (3.58)
eADHD$_{integrated}$	11.55 (4.24)	6.89 (6.02)	6.79 (5.70)

Note. %SS = percentage of syllables stuttered; No-eADHD$_{standard}$ = children who stutter without elevated ADHD symptoms who received stuttering treatment only; eADHD$_{standard}$ = children who stutter with elevated ADHD symptoms who received stuttering treatment only; eADHD$_{integrated}$ = children who stutter with elevated ADHD symptoms who received the integrated self-regulation and stuttering treatment program; ADHD = attention-deficit/hyperactivity disorder; SDQ total = total scale score from the Strengths and Difficulties Questionnaire (R. Goodman, 1997); CBQ-EC = effortful control domain on the Child Behavior Questionnaire (Rothbart et al., 2001); PAFAS = Parenting and Family Adjustment Scales (Sanders, Morawska, et al., 2014; lower scores on the PAFAS scales indicate more desirable parenting practices and family adjustment).

[a]No-eADHD$_{standard}$, *n* = 40; eADHD$_{standard}$, *n* = 17; eADHD$_{integrated}$, *n* = 19. [b]No-eADHD$_{standard}$, *n* = 40; eADHD$_{standard}$, *n* = 16; eADHD$_{integrated}$, *n* = 19. [c]No-eADHD$_{standard}$, *n* = 39; eADHD$_{standard}$, *n* = 16 for %SS and *n* = 14 for the remaining measures (due to nonreturned questionnaires); eADHD$_{integrated}$, *n* = 19.

group and time, $F(1, 67) = 27.32$, $p < .001$, $\eta^2 = .44$. As shown in Table 3, the interaction appears to be due to a larger pre- to postreduction in ADHD-IV combined scores for the eADHD$_{integrated}$ compared to the eADHD$_{standard}$ group. Simple effects support this interpretation and hypotheses made: The pre- to posttreatment contrast was significant for the eADHD$_{integrated}$ group, $t(67) = 9.78$, $p < .001$, but not the eADHD$_{standard}$ group, $t(67) = 0.86$, $p = .394$, and at posttreatment, the ADHD-IV combined scores for the eADHD$_{integrated}$ group were significantly lower than those for the eADHD$_{standard}$ group, $t(67) = 2.73$, $p = .008$.

A similar pattern of results was obtained for the analysis of pretreatment and follow-up ADHD-IV combined scores. There was no significant effect of group, $F < 1$, but

there was a significant effect of time, $F(1, 65) = 65.70$, $p < .001$, $\eta^2 = .65$, and a Group × Time interaction, $F(1, 65) = 25.70$, $p < .001$, $\eta^2 = .45$. The change from pretreatment to follow-up in ADHD-IV combined scores was larger for the eADHD$_{integrated}$ group compared to the eADHD$_{standard}$ group (see Table 3). Comparisons of marginal means showed the effect of time was significant for the eADHD$_{integrated}$ group, $t(65) = 10.46$, $p < .001$, and was bordering on significant for the eADHD$_{standard}$ group, $t(65) = 1.95$, $p = .056$. This suggests some reduction in ADHD symptomatology over time (i.e., at follow-up) for the eADHD$_{standard}$ group, although this was not as large as the change observed for the eADHD$_{integrated}$ group. At follow-up, the eADHD$_{integrated}$ group was significantly lower in ADHD combined scores

Figure 2. Mean percent syllables stuttered at pretreatment, posttreatment, and follow-up for the children who stutter with elevated attention-deficit/hyperactivity disorder (ADHD) symptoms receiving the standard stuttering treatment (eADHD$_{standard}$) and children who stutter with elevated ADHD symptoms receiving the integrated treatment (eADHD$_{integrated}$) along with 95% confidence intervals.
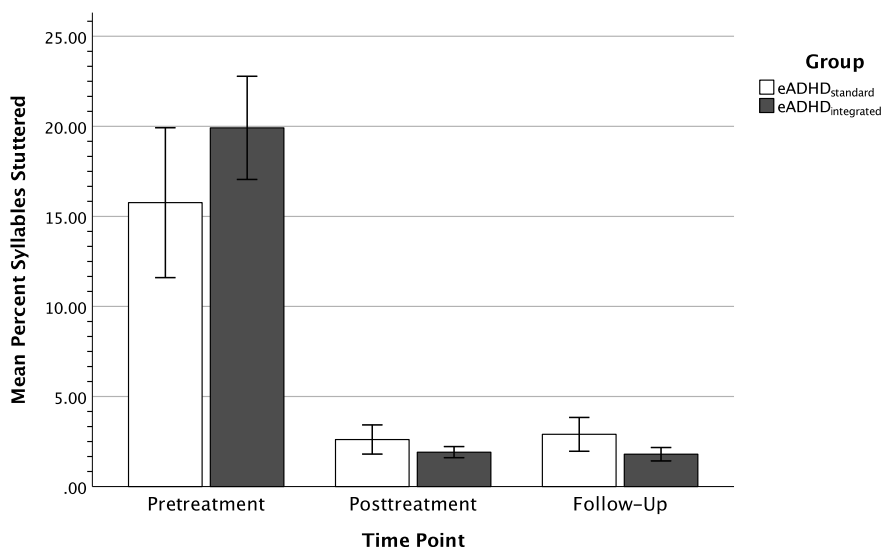
than the eADHD$_{standard}$ group, $t(65) = 2.78$, $p = .007$. The pattern of results we observed in the ADHD-IV combined scores was also observed in the ADHD-IV inattentive and ADHD-IV hyperactive/impulsive subscale data.

SDQ

The GLMM comparing eADHD$_{integrated}$ and eADHD$_{standard}$ groups in their pre- and posttreatment SDQ scores showed a significant effect of group, $F(1, 67) = 4.64$, $p = .035$, $\eta^2 = .09$, and time, $F(1, 67) = 37.61$, $p < .001$, $\eta^2 = .51$, as well as an interaction between group and time, $F(1, 67) = 27.57$, $p < .001$, $\eta^2 = .43$. As shown in Table 3, similar to the ADHD combined scores, the interaction appears to be due to a larger pre- to postreduction in SDQ scores for the eADHD$_{integrated}$ compared to the eADHD$_{standard}$ group. Marginal mean comparisons once again support this interpretation and are in line with our hypotheses: The pre- to posttreatment contrast was significant for the eADHD$_{integrated}$ group, $t(67) = 8.35$, $p < .001$, but not the eADHD$_{standard}$ group, $t(67) = 0.60$, $p = .135$. At pretreatment, the eADHD$_{integrated}$ group showed significantly higher SDQ scores than the eADHD$_{standard}$ group. At posttreatment, the eADHD$_{integrated}$ group SDQ scores were lower, although this difference was not statistically significant, $t(67) = 1.11$, $p = .269$.

A similar pattern of results was obtained for the analysis of pretreatment and follow-up SDQ scores. There was a significant effect of group, $F(1, 65) = 6.02$, $p = .017$, $\eta^2 = .11$, and time, $F(1, 65) = 42.51$, $p < .001$, $\eta^2 = .53$, and a Group × Time interaction, $F(1, 65) = 22.18$, $p < .001$, $\eta^2 = .36$. Comparisons of marginal means showed the effect of time was significant for the eADHD$_{integrated}$ group, $t(65) = 7.48$, $p < .001$, and nonsignificant for the eADHD$_{standard}$

group, $t(65) = 1.37$, $p = .175$, showing a larger reduction in SDQ scores for the eADHD$_{integrated}$ group. There was no significant difference between groups at follow-up, $t(65) = 0.56$, $p = .563$, which contrasts with pretreatment comparisons where the eADHD$_{integrated}$ group was significantly higher in SDQ total scores.

CBQ-EC

The GLMM comparing the eADHD$_{integrated}$ and eADHD$_{standard}$ groups in their pre- and posttreatment CBQ-EC scores showed nonsignificant effects for group, $F < 1$, but a significant effect of time, $F(1, 67) = 10.45$, $p = .002$, $\eta^2 = .23$, and a significant interaction between group and time, $F(1, 67) = 4.30$, $p = .042$, $\eta^2 = .11$.

As shown in Table 3, the interaction appears to be due to a larger pre- to post increase in CBQ-EC for the eADHD$_{integrated}$ compared to the eADHD$_{standard}$ group. As predicted, the pre- and post difference in CBQ-EC scores was significant for the eADHD$_{integrated}$ group, $t(67) = 4.22$, $p < .001$, but not the eADHD$_{standard}$ group, $t(67) = 0.74$, $p = .459$.

For the analysis of pretreatment and follow-up CBQ-EC scores, there was no significant effect of group, $F > 1$, but time was significant, $F(1, 65) = 16.78$, $p < .001$, $\eta^2 = .35$. The Group × Time interaction, however, was not significant, $F(1, 65) = 3.43$, $p = .069$, $\eta^2 = .11$, suggesting the increase from pretreatment to follow-up in CBQ-EC scores, while numerically larger for the eADHD$_{integrated}$ group, was not significantly larger.

PAFAS Parenting Practices

The GLMM comparing the eADHD$_{integrated}$ and eADHD$_{standard}$ groups in their pre- and posttreatment

PAFAS parenting practices scores showed no significant effect for group, $F < 1$, but a significant effect of time, $F(1, 67) = 47.22$, $p < .001$, $\eta^2 = .57$, as well as an interaction between group and time, $F(1, 67) = 51.21$, $p < .001$, $\eta^2 = .59$. As shown in Table 3, the interaction appears to be due to a larger pre- to postreduction in scores for the eADHD$_{integrated}$ compared to the eADHD$_{standard}$ group. As predicted, and in support of this interpretation, the pre- to posttreatment contrast was significant for the eADHD$_{integrated}$ group, $t(67) = 10.55$, $p < .001$, but not the eADHD$_{standard}$ group, $t(67) = 0.19$, $p = .850$. At pretreatment, the eADHD$_{integrated}$ group showed significantly higher (less desirable) parenting practices scores than the eADHD$_{standard}$ group, but significantly lower (more desirable) scores at posttreatment, $t(67) = 3.21$, $p = .002$ (see Table 3).

A similar pattern of results was obtained for the analysis of pretreatment and follow-up PAFAS parenting practices scores. There was no significant effect for group, $F < 1$, but a significant effect of time, $F(1, 65) = 25.38$, $p < .001$, $\eta^2 = .40$, as well as an interaction between group and time, $F(1, 64) = 36.41$, $p < .001$, $\eta^2 = .51$. Comparisons of marginal means showed the effect of time was significant for the eADHD$_{integrated}$ group, $t(65) = 8.15$, $p < .001$, and nonsignificant for the eADHD$_{standard}$ group, $t(65) = 0.68$, $p = .500$. In addition, the eADHD$_{integrated}$ group demonstrated significantly lower scores than the eADHD$_{standard}$ group at follow-up, $t(65) = 3.49$, $p = .001$ (see Table 3).

PAFAS Family Adjustment

The GLMM comparing the eADHD$_{integrated}$ and eADHD$_{standard}$ groups in their pre- and posttreatment PAFAS family adjustment scores showed no significant effects for group, $F < 1$, and time, $F < 1$, but a significant Group × Time interaction, $F(1, 67) = 28.02$, $p < .001$, $\eta^2 = .437$. The interaction appears to be due to a predicted and significant pre- to postreduction in scores for the eADHD$_{integrated}$ group, $t(67) = 4.57$, $p < .001$, and, unexpectedly, a significant increase in scores in the eADHD$_{standard}$ group, $t(67) = 2.89$, $p = .005$. The eADHD$_{integrated}$ group showed significantly lower posttreatment scores compared to the eADHD$_{standard}$ group, $t(67) = 3.03$, $p < .01$.

Similarly, for the analysis of pretreatment and follow-up PAFAS family adjustment scores, there were no significant effects for group, $F > 1$, and time, $F > 1$, but a significant Group × Time interaction, $F(1, 64) = 21.81$, $p < .001$, $\eta^2 = .433$. The effect of time was significant for the eADHD$_{integrated}$ group, $t(64) = 2.46$, $p = .016$, but not significant for the eADHD$_{standard}$ group, $t(64) = 0.32$, $p = .318$. The eADHD$_{integrated}$ group showed significantly lower (i.e., more desirable) scores at follow-up than the eADHD$_{standard}$ group, $t(64) = 2.37$, $p = .021$.

No-eADHD Group Outcomes and Comparisons With eADHD Groups (Secondary Aims c and d)

Outcomes for the No-eADHD$_{standard}$ group will be considered first (see Table 3). When compared to pretreatment, %SS was significantly lower for the No-eADHD$_{standard}$

group at posttreatment, $t(79) = 9.13$, $p < .001$, and at follow-up, $t(79) = 9.45$, $p < .001$. Only two of 41 (5%) CWS without comorbid ADHD symptoms did not reach the study's discharge criterion at follow-up. These results provide further support for the CUSP as an effective early intervention program for preschool CWS, in line with our hypotheses.

Regarding psychosocial outcomes for the No-eADHD$_{standard}$ group, there were no significant changes from pre to post or from pre to follow-up for ADHD symptoms, SDQ total, and PAFAS parenting practices and family adjustment scores ($ps > .05$). There was, however, a significant increase in the No-eADHD$_{standard}$ group's CBQ-EC scores from pretreatment to posttreatment, $t(79) = 3.00$, $p = .004$, and from pretreatment to follow-up, $t(78) = 4.18$, $p < .001$.

To align with the secondary study aim relating to the comparison of presentation characteristics between CWS with and without eADHD, GLMM analyses compared the No-eADHD$_{standard}$ group to the eADHD groups on outcome measures at pretreatment, posttreatment, and follow-up. Consistent with the hypotheses, the No-eADHD$_{standard}$ group compared to the eADHD group as a whole (combined eADHD$_{integrated}$ and eADHD$_{standard}$ children) showed significantly lower pretreatment %SS scores, $t(75) = 3.76$, $p < .001$, and significantly higher CBQ-EC scores, $t(75) = 5.12$, $p < .001$. In addition, SDQ scores were expectedly significantly lower in this group, $t(75) = 8.40$, $p < .001$.

After treatment, the eADHD$_{standard}$ group remained higher than the No-eADHD$_{standard}$ group in the primary outcome measure, %SS. This difference did not reach statistical significance at posttreatment, $t(72) = 2.36$, $p = .063$, but did at follow-up, $t(72) = 3.27$, $p = .005$. There was no significant difference in %SS between the eADHD$_{integrated}$ and No-eADHD$_{standard}$ groups at posttreatment, $t(72) = 1.13$, $p = .260$, or follow-up, $t(72) = 1.63$, $p = .107$.

With number of treatment sessions as the outcome, LSD contrasts (after controlling for differences in %SS) showed the No-eADHD$_{standard}$ group (adjusted $M = 10.2$, $SEM = 0.6$) required significantly fewer stuttering treatment sessions to discharge compared to the eADHD$_{standard}$ group, $t(71) = 2.73$, $p = .024$, but there was no significant difference in number of stuttering treatment sessions when compared to the eADHD$_{integrated}$ group, $t(71) = 0.32$, $p = .752$.[3]

With respect to the psychosocial or secondary outcome measures, when compared to the No-eADHD$_{standard}$ group at posttreatment and follow-up, the eADHD$_{integrated}$ and eADHD$_{standard}$ groups remained significantly higher in ADHD-IV scores and lower in CBQ-EC scores ($ps < .008$). The eADHD$_{standard}$ group remained significantly higher in SDQ scores than the No-eADHD$_{standard}$ group at posttreatment and follow-up ($ps < .002$), while the eADHD$_{integrated}$ group was only significantly higher at follow-up ($p = .047$). For the PAFAS parenting practices, the eADHD$_{integrated}$ group was significantly lower than both the No-eADHD$_{standard}$ and eADHD$_{standard}$ groups at posttreatment and follow-up

---

[3]The unadjusted mean for the No-eADHD is 9.7 ($SD = 3.6$).

(*p*s < .01). Finally, for the PAFAS family adjustment scores, the eADHD$_{standard}$ group was significantly higher than the No-eADHD$_{standard}$ group at posttreatment and follow-up (*p*s < .001), whereas there was no difference between the eADHD$_{integrated}$ and No-eADHD$_{standard}$ groups at both of those time points (*p*s > .364).

Discussion

Fluency Outcomes and Stuttering Treatment Responsiveness

A significant overall reduction in stuttered speech severity was obtained by the end of the treatment for all treatment groups, with improvements successfully maintained at follow-up. This provides evidence that the CUSP, which combines direct and indirect fluency management strategies to facilitate naturalistic and individualized stuttering intervention (Druker, Mazzucchelli, & Beilby, 2019), was successful for this large group of preschool-age CWS. The attrition rate of 1.32% at posttreatment, and a cumulative 5.26% at follow-up, was very low in comparison to previously published attrition rates for preschool stuttering interventions (McCulloch et al., 2017). This may be due in part to the nature of the fluency intervention, whereby stuttering treatment principles align with general parenting principles and are flexibly embedded throughout daily parent–child interactions (Druker, Mazzucchelli, & Beilby, 2019). They are naturalistic in form and do not require designated practice activities or structured materials.

It is worthy to note the 25% nonresponse rate for children in the eADHD$_{standard}$ group is equivalent to the nonresponse rate documented in a recent randomized controlled trial of stuttering intervention, providing further support for the CUSP as an effective early intervention (de Sonneville-Koedoot et al., 2015). The very low nonresponse rate (5%) in the integrated group, however, underscores the importance and successful nature of addressing comorbid factors in early stuttering disorders to maximize recovery (Donaghy & Smith, 2016).

Consistent with our primary hypothesis, children who received the integrated program demonstrated significantly lower stuttered speech severity at follow-up compared to the eADHD$_{standard}$ group. Findings from this study suggest that addressing self-regulation skills facilitates enhanced stuttering treatment outcomes, which in turn is supportive of research proposing that stuttering has emotional regulation aspects that also require management (Walden et al., 2012). These findings also endorse future clinical implementation of this integrated program for a cohort of CWS with self-regulation challenges, who typically respond less successfully to stuttering treatment (Druker, Hennessey, et al., 2019; Riley & Riley, 2000).

In the context of increased demands on community health care resources, the consideration of the cost-effectiveness of health interventions is important (Kernick, 2003). In this light, it is worthy to compare the benefits gained by including EBPS in stuttering interventions for a cohort of CWS

with self-regulation challenges. Previously published early stuttering interventions report averages of up to 19.6 hr of clinical intervention time (e.g., de Sonneville-Koedoot et al., 2015). In the current study, the average duration of a stuttering intervention session was 40 min, with a mean number of 11 sessions for the group of children receiving the integrated program (a total of 7.3 hr). The additional EBPS intervention time was 5.75 hr, resulting in total intervention time of approximately 12.08 hr.

As predicted, the children in this study with eADHD symptoms whose parents received EBPS required fewer face-to-face stuttering treatment sessions compared to those who did not receive EBPS. This finding is of interest because it not only shows the additional EBPS time can potentially be offset to some degree by fewer stuttering treatment sessions, but it also suggests responsiveness to a stuttering treatment program focused primarily on managing the stuttering behavior can be improved through the indirect effects of supporting parents to enhance their child's self-regulation. A corollary of this point is that, at least for children with eADHD symptoms, successful treatment of stuttering was achieved not just through treatment components that target the stuttering behavior.

Furthermore, the EBPS component of the intervention may support the longer term maintenance of stuttering outcomes given the significant and sustained improvement in the children's self-regulations skills (Eggers et al., 2013; Karrass et al., 2006; Walden et al., 2012). In addition, the promotion of positive parent–child interactions, as well as improved predictability and positivity within the home environment reported as a result of the successful Triple P component of the integrated program (Starkweather, 1987), is likely to indirectly support these children in their long-term capacity for fluency, already evidenced by their enhanced stuttering outcomes at 3 months of follow-up. Further longitudinal follow-up of these children would be of value to investigate the longer term impact of this integrated program. The additional benefits likely derived from receiving the EBPS content are important to consider, given the possible psychosocial consequences of eADHD symptoms for CWS and their parents, if not successfully addressed in the early formative years (Druker, Mazzucchelli, & Beilby, 2019; Mazzucchelli, 2018; Mazzucchelli & Sanders, 2018).

A future research direction could be to match intervention time, where one group receives the integrated intervention and the other group receives the same amount of intervention, but all devoted to stuttering. This future research will test predictions made above relating to EBPS playing a crucial role in maintaining long-term fluency benefits.

Psychosocial Outcomes for CWS

Children who received the integrated program demonstrated a significant pre–post reduction in inattentive and hyperactive/impulsive subscale scores, as well as the overall combined ADHD symptom score, on the ADHD Rating Scale-IV. Furthermore, a significant reduction in the

total difficulties score of the SDQ was also found for these children, which is a recognized measure of child mental health (A. Goodman & Goodman, 2012). These changes were maintained over 3 months posttreatment. No pre–post changes on these measures occurred in the control eADHD group, supporting the internal validity of these significant findings.

Although CWS did not necessarily meet criteria for a clinical ADHD diagnosis, a marked positive shift in both ADHD symptoms and scores measuring overall socioemotional well-being augers well for potential long-term benefits for CWS in the eADHD$_{integrated}$ group (Duckworth & Seligman, 2005; Moffitt et al., 2011; Tsukayama et al., 2010). In addition, significant improvements in effortful control were unexpectedly evident at posttreatment for CWS who received the integrated program, with a trend in a similar direction at follow-up. The No-ADHD$_{standard}$ group also showed improvement in effortful control after treatment. Effortful control is an aspect of self-regulation that refers to the "ability to inhibit a dominant response and execute a subdominant response" (Anzman-Frasca et al., 2015, p. 204). Robust evidence links this specific component of self-regulation to multiple aspects of physical and mental well-being, including social competence and adjustment, academic outcomes, and child weight status trajectory (Eisenberg et al., 2005; Francis & Susman, 2009; NSCAW, 2009; Smith-Donald et al., 2007). Therefore, the positive shift in effortful control found in the eADHD$_{integrated}$ group of children at posttreatment, in particular, provides preliminary support for Triple P as a successful parent intervention to facilitate this particular characteristic of child self-regulation improvement, at least in the short term. It also suggests that successful treatment for stuttering in children who are not compromised by behavioral difficulties may contribute to improvements in self-regulation.

Eggers et al. (2013) maintain that weaker self-regulation skills may increase the emotional arousal experienced by some CWS, thus exacerbating their stuttering symptoms. These authors recommend that supporting children to develop self-regulation skills may decrease emotional arousal and thus reduce its impact on stuttering symptoms. Furthermore, they extend the possibility of reduced self-regulatory capacity to be linked to a tendency for persistent stuttering. The current study is the first study to successfully demonstrate improved self-regulatory behaviors in CWS, with possible future implications for enhanced prognostic outcomes in the stuttering trajectories of these young children.

Psychosocial Outcomes for Parents

Parents have been shown to be key agents of change in contemporary stuttering interventions (de Sonneville-Koedoot et al., 2015; Druker, Mazzucchelli, Hennessey, & Beilby, 2019; Franken & Putker-de Bruijn, 2007; Millard et al., 2008; Onslow & O'Brian, 2013; Yaruss et al., 2006). Furthermore, parents of CWS have been found to experience negative emotional reactions, difficulty coping, anxiety, and perceived lack of support in response to their child's overall stuttering (Druker, Mazzucchelli, & Beilby, 2019; Langevin et al., 2010). This underscores the importance of empowering parents to confidently and holistically manage all aspects of the child's needs, particularly those aspects that have been found to impact stuttering treatment responsiveness and outcomes.

The improved parenting practices of parents who received the integrated program, consistent with the study's hypothesis, indicate that these parents were likely confident and consistent in managing their children's stuttering overall, although this was not formally assessed. The overall improvement in family adjustment indicates that parents of CWS who received the integrated intervention were coping better with their parental role, and their families were functioning more cohesively as an integrated unit (Sanders, Morawska, et al., 2014). Beyond the significant benefits of these findings on parent and child mental health, findings can also be considered in light of recent qualitative research that highlighted how cohesive families do better in managing stuttering and related sequelae (Druker, Mazzucchelli, & Beilby, 2019). Once again, in line with the study's predictions, no change in parenting practices occurred in the control eADHD group or in the children with no eADHD symptoms whose stuttering treatment was also successful. There was an unexpected pre- to postincrease (i.e., less desirable change) in family adjustment scores in the eADHD$_{standard}$ group; however, this difference was not maintained at follow-up. The significant finding in the pre–post comparison is consistent with literature highlighting the link between parenting stress and child behavior problems (Neece et al., 2012).

It is worthwhile highlighting that CWS in the eADHD$_{integrated}$ group had significantly greater ADHD Rating Scale-IV scores at pretreatment, compared to the eADHD$_{standard}$ group. The large reduction in ADHD symptom scores in the eADHD$_{integrated}$ group resulted in these children presenting with significantly lower ADHD scores at posttreatment, compared to those in the eADHD$_{standard}$. This impressive pattern was similar for parenting practices, whereby the eADHD$_{integrated}$ group presented with poorer parenting practices at pretreatment, and significantly more desirable parenting practices at posttreatment, compared to the eADHD$_{standard}$ group. In short, the children and their parents in this group can be seen to have benefited substantially from their participation in this research.

To address the study's additional aim of comparing characteristics of CWS with and without ADHD symptoms, pretreatment stuttered speech severity and behavioral measures were compared to all CWS with elevated symptoms, regardless of their treatment group classification. In line with our hypotheses, CWS without elevated symptoms presented with significantly lower %SS at pretreatment compared to CWS who met criteria for eADHD. This supports previous research that identified CWS with higher effortful control to have less severe stuttered speech severity at pretreatment (and vice versa) and also supports the claim that CWS with eADHD have difficulties with self-regulation (Kraft et al., 2014, 2019). Finally, in further support of

CWS with eADHD having difficulties with self-regulation, as predicted, CWS without eADHD presented with significantly lower SDQ scores and higher effortful control than the other two groups who met criteria for eADHD.

The posttreatment stuttered speech severity was enhanced in the No-eADHD group compared to CWS with eADHD, in line with our hypotheses. Following further analysis, however, it became evident that posttreatment %SS in the No-eADHD group was similar to that of CWS who received the integrated program (and significantly lower than the eADHD$_{standard}$ group only). Ultimately, these findings provide theoretical and clinical support for the integrated treatment in this study, which has elucidated how concurrent management of self-regulation skills supports CWS challenged with self-regulation difficulties to successfully allocate resources to ongoing fluent speech production. In doing so, the possible impact of these comorbid challenges on stuttering treatment outcomes is mitigated, resulting in posttreatment success and positive follow-up outcomes similar to those of CWS without the concomitant symptoms.

Limitations and Future Directions

The EBPS program included examples of positive parenting practices (e.g., using praise) to help CWS regulate their stuttering behavior and other behaviors. It is unclear, therefore, whether general improvements in the eADHD$_{integrated}$ children's self-regulation capacity, as demonstrated by improved effortful control, were responsible for the positive effect of the integrated treatment, including improved treatment responsiveness, or the fact that the program also focused on self-regulating stuttering behaviors. These accounts could potentially be teased apart in future research by comparing the current integrated version of the Triple P with a version of the program that has not been adapted to stuttering.

The aim of this study was to evaluate the effectiveness of the integrated program for CWS with subclinical levels of ADHD. Therefore, findings may not be generalizable to CWS with clinically diagnosed ADHD, as CWS with clinical ADHD may be ultimately more resistant to treatment. Future research is warranted to evaluate the effectiveness of a similar integrated self-regulation and stuttering treatment program for CWS with clinical ADHD diagnoses, to determine if the program yields similar positive outcomes.

Of the parents who had children with eADHD, 81% were randomized to the two intervention conditions. The nonrandom assignment of 19% of participants has limitations in terms of generalizing the relative efficacy of the two intervention conditions. Parents opting to receive the program were likely to be experiencing greater challenges with their parenting practices and their child's behavior, and therefore, these parents may have been more receptive to receiving combined stuttering and parenting support. This possibility also needs to be considered in view of possible reporting bias influencing parent report measures of child behavior and parenting outcomes. Given the significance of findings yielded through parent report measures, it is recommended that future research include observational measures to augment objectivity of findings and eliminate possible bias inherent in parent report measures (Hawes & Dadds, 2006).

Despite 81% of CWS with eADHD being randomized to group, statistically significant differences existed at pretreatment between these two groups on key outcome variables, including ADHD Rating Scale-IV, SDQ, and PAFAS scores. Between-group differences at pretreatment may limit the internal validity of study findings. However, these differences were unlikely to bias the results because there were significantly lower posttreatment scores for families in the eADHD$_{integrated}$ group compared to the families in the eADHD$_{standard}$, despite scores in the eADHD$_{integrated}$ being significantly higher at pretreatment.

This partial nonrandom assignment may have contributed to the significant differences in child behavior and parenting practices across groups at pretreatment. This supports the notion that this integrated intervention may be most suitable for CWS with eADHD who have parents requiring additional parenting support. Future research and clinical implementation may consider a pretreatment parent screening protocol to test for suitability, before recommending this integrated intervention program.

The organizational nature of the CSTC and affiliated off-site clinic around the academic timetable of the university did not allow for effects of site to be analyzed independently; however, the children received the same intervention, at both clinics, at some point in their treatment journey. This is due to student placement schedules and the CSTC demands requiring all students to gain experience in managing early developmental stuttering disorders with treatment continuing during the student vacation periods. Although this is a strength in terms of the clinical validity of the treatment, future research may consider separating CWS between treatment sites to allow effects of site to be analyzed in treatment outcomes.

Spontaneous recovery was not controlled for when evaluating treatment outcomes. However, given the ambiguity of research surrounding natural recovery and the very low recovery rate 12 months after onset (Reilly et al., 2013), clinics from which participants were recruited promote the importance of and subscribe to early stuttering intervention as close as possible to stuttering onset.

Finally, authors recommend the use of additional psychosocial measures in future developmental stuttering treatment research, to capture possible changes in emotions and communicative attitudes associated with improved speech fluency in this population (e.g., Vanryckeghem et al., 2005).

Conclusions

The importance of addressing all aspects of a child's stuttered speech disorder and any associated behavioral self-regulatory challenges has been highlighted in contemporary literature (Eggers et al., 2013; Smith & Weber, 2017). This study implemented an integrated stuttering and self-regulation management program for CWS with eADHD symptoms.

The findings demonstrate that employing an EBPS program in conjunction with an early stuttering treatment was successful for CWS with eADHD and their parents, in terms of improving fluency outcomes, responsiveness to stuttering treatment, and child and parent psychosocial outcomes.

Acknowledgments

References

Alm, P. A., & Risberg, J. (2007). Stuttering in adults: The acoustic startle response, temperamental traits, and biological factors. *Journal of Communication Disorders, 40*(1), 1–41. https://doi.org/10.1016/j.jcomdis.2006.04.001

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.).

Anzman-Frasca, S., Francis, L., & Birch, L. (2015). Inhibitory control is associated with psychosocial, cognitive, and weight outcomes in a longitudinal sample of girls. *Psychological Science, 1*(3), 203–216. https://doi.org/10.1037/tps0000028

Barkley, R. (2013). *Taking charge of ADHD: The complete, authoritative guide for parents* (3rd ed.). The Guilford Press.

Beilby, J. (2014). Psychosocial impact of living with a stuttering disorder: Knowing is not enough. *Seminars in Speech and Language, 35*(2), 132–143. https://doi.org/10.1055/s-0034-1371756

Bjornebekk, G., Kjobli, J., & Ogden, T. (2015). Children with conduct problems and co-occurring ADHD: Behavioral improvements following parent managment training. *Child & Family Behavior Therapy, 37*(1), 1–19. https://doi.org/10.1080/07317107.2015.1000227

Bor, W., Sanders, M., & Markie-Dadds, C. (2002). The effects of the Triple P-Positive Parenting Program on preschool children with co-occurring disruptive behavior and attentional/hyperactive difficulties. *Journal of Abnormal Child Psychology, 30*(6), 571–587. https://doi.org/10.1023/A:1020807613155

Bradshaw, S., Bem, D., Shaw, K., Taylor, B., Chiswell, C., Salama, M., Bassett, E., KAur, G., & Cummins, C. (2019). Improving health, wellbeing and parenting skills in parents of children with special health care needs and medical complexity—A scoping review. *BMC Pediatrics, 19*(301), 1–11. https://doi.org/10.1186/s12887-019-1648-7

Clark, C., Prior, M., & Kinsella, G. (2002). The relationship between executive function abilities, adaptive behavior and academic achievment in children with externalizing behavior problems. *The Journal of Child Psychology and Psychiatry, 43*(6), 785–796. https://doi.org/10.1111/1469-7610.00084

de Sonneville-Koedoot, C., Stolk, E., Rietveld, T., & Franken, M.-C. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PLOS ONE, 10*(7), 1–17. https://doi.org/10.1371/journal.pone.0133758

Dodd, B., Hua, Z., Crosbie, S., Holm, A., & Ozanne, A. (2002). *Diagnostic evaluation of articulation and phonology*. The Psychological Corporation.

Donaghy, M., & Smith, K. (2016). Management options for pediatric patients who stutter: Current challenges and future directions. *Pediatric Health, Medicine and Therapeutics, 7,* 71–77. https://doi.org/10.2147/PHMT.S77568

Donaher, J., & Richels, C. (2012). Traits of attention deficit/hyperactivity disorder in school-age children who stutter. *Journal of Fluency Disorders, 37*(4), 242–252. https://doi.org/10.1016/j.jfludis.2012.08.002

Druker, K., Hennessey, N., Mazzucchelli, T. G., & Beilby, J. (2019). Elevated attention deficit hyperactivity disorder symptoms in children who stutter. *Journal of Fluency Disorders, 59,* 80–90. https://doi.org/10.1016/j.jfludis.2018.11.002

Druker, K., Mazzucchelli, T., Hennessey, N., & Beilby, J. (2018). *Triple P and stuttering tip sheet*. Curtin University.

Druker, K., Mazzucchelli, T., Hennessey, N., & Beilby, J. (2019). Parent perceptions of an integrated stuttering treatment and behavioral self-regulation program for early developmental stuttering. *Journal of Fluency Disorders, 62,* 1–17. https://doi.org/10.1016/j.jfludis.2019.105726

Druker, K., Mazzucchelli, T. G., & Beilby, J. (2019). An evaluation of integrated fluency and resilience program for early developmental stuttering disorders. *Journal of Communication Disorders, 78,* 69–83. https://doi.org/10.1016/j.jcomdis.2019.02.002

Duckworth, A. L., & Seligman, M. (2005). Self-discipline outdoes IQ in predicting academic performance in adolescents. *Psychological Science, 16*(12), 939–944. https://doi.org/10.1111/j.1467-9280.2005.01641.x

DuPaul, G. J., Anastopoulos, A. D., Power, T. J., Reid, R., Ikeda, M. J., & McGoey, K. E. (1998). Parent ratings of attention-deficit/hyperactivity disorder symptoms: Factor structure and normative data. *Journal of Psychopathology and Behavioral Assessment, 20,* 83–102. https://doi.org/10.1023/A:1023087410712

Eggers, K., De Nil, L., & Van den Bergh, B. (2010). Temperament dimensions in stuttering and typically developing children. *Journal of Fluency Disorders, 35*(4), 355–372. https://doi.org/10.1016/j.jfludis.2010.10.004

Eggers, K., De Nil, L., & Van Den Bergh, B. (2013). Inhibitory control in childhood stuttering. *Journal of Fluency Disorders, 38*(1), 1–13. https://doi.org/10.1016/j.jfludis.2012.10.001

Eisenberg, N., & Spinrad, T. (2004). Emotion-related regulation: Sharpening the definition. *Child Development, 75*(2), 334–339. https://doi.org/10.1111/j.1467-8624.2004.00674.x

Eisenberg, N., & Sulik, M. J. (2012). Emotion-related self-regulation in children. *Teaching of Psychology, 39*(1), 77–83. https://doi.org/10.1177/0098628311430172

Eisenberg, N., Zhou, Q., Spinrad, T., Valiente, C., Fabes, R., & Liew, J. (2005). Relations among positive parenting, children's effortful control, and externalizing problems: A three-wave longitudinal study. *Journal of Child Development, 76*(5), 1055–1071. https://doi.org/10.1111/j.1467-8624.2005.00897.x

Felsenfeld, S., Van Beljsterveldt, C., & Boomsma, D. I. (2010). Attentional regulation in young twins with probable stuttering, high nonfluency, and typical fluency. *Journal of Speech, Language, and Hearing Research, 53*(5), 1147–1166. https://doi.org/10.1044/1092-4388(2010/09-0164)

ForFluency. (2012). *Fluency rater*. [Mobile app]. Download.com. http://download.cnet.com/Fluency-Rater/3000-2129_4-75357634.html

Francis, L., & Susman, E. (2009). Self-regulation and rapid weight gain in children from age 3 to 12 years. *Archives of Pediatrics & Adolescent Medicine, 163*(4), 297–302. https://doi.org/10.1001/archpediatrics.2008.579

Franken, M. C., & Putker-de Bruijn, D. (2007). *Restart-DCM method. Treatment protocol developed within the scope of the*

ZonMW project: Cost-effectiveness of the Demands and Capacities Model based treatment compared to the Lidcombe programme of early stuttering intervention: Randomised trial (K. Laird, Trans.). Erasmus MC.

Fujiwara, T., Kato, N., & Sanders, M. (2011). Effectiveness of Group Positve Parenting Program (Triple P) in changing child behavior, parenting style, and parental adjustment: An intervention study in Japan. *Journal of Child and Family Studies, 20*(6), 804–813. https://doi.org/10.1007/s10826-011-9448-1

Goodman, A., & Goodman, R. (2012). Strengths and Difficulties Questionnaire scores and mental health in looked after children. *The British Journal of Psychiatry, 200*(5), 426–427. https://doi.org/10.1192/bjp.bp.111.104380

Goodman, R. (1997). The Strengths and Difficulties Questionnaire: A research note. *The Journal of Child Psychology and Psychiatry, 38,* 581–586. https://doi.org/10.1111/j.1469-7610.1997.tb01545.x

Goodman, R., Ford, T., Simmons, H., Gatward, R., & Meltzer, H. (2000). Using the Strengths and Difficulties Questionnaire (SDQ) to screen for child psychiatric disorders in a community sample. *British Journal of Psychiatry, 117*(6), 534–539. https://doi.org/10.1192/bjp.177.6.534

Goodman, R., Renfrew, D., & Mullick, M. (2000). Predicting type of psychiatric disorder from Strengths and Difficulties Questionnaire (SDQ) scores in child mental health clinics in London and Dhaka. *European Child & Adolescent Psychiatry, 9,* 129–134. https://doi.org/10.1007/s007870050008

Grey, M., Goodrich, A., Jaser, S., & Lindemann, E. (2011). Coping skills traning for parents of children with Type 1 diabetes: 12-month outcomes. *Nursing Research, 60*(3), 173–181. https://doi.org/10.1097/NNR.0b013e3182159c8f

Guitar, B. (2014). *Stuttering: An integrated approach to its nature and treatment* (4th ed.). Lippincott Williams & Wilkins.

Hawes, D., & Dadds, M. (2006). Assessing parenting practices through parent report and direct observation during parent training. *Journal of Child and Family Studies, 15,* 554–567. https://doi.org/10.1007/s10826-006-9029-x

Hoath, F., & Sanders, M. (2002). A feasibility study of Enhanced Group Triple P—Positive parenting program for parents of children with Attention-deficit/hyperactivity disorder. *Behaviour Change, 19*(4), 191–206. https://doi.org/10.1375/bech.19.4.191

Huguet, A., Eguren, J., Miguel-Ruiz, D., Valles, X., & Alda, A. (2019). Deficient emotional self-regulation in children with attention deficit hyperactivity disorder: Mindfulness as a useful treatment modality. *Journal of Developmental & Behavioral Pediatrics, 40*(6), 425–431. https://doi.org/10.1097/DBP.0000000000000682

Jones, R., Choi, D., Conture, E., & Walden, T. (2014). Temperament, emotion and childhood stuttering. *Seminars in Speech and Language, 35*(2), 114–131. https://doi.org/10.1055/s-0034-1371755

Karimi, H., Jones, M., O'Brian, S., & Onslow, M. (2014). Clinician percent syllables stuttered, clinician severity ratings and speaker severity ratings: Are they interchangeable? *International Journal of Language & Communication Disorders, 49*(3), 364–368. https://doi.org/10.1111/1460-6984.12069

Karrass, J., Walden, T., Conture, E., Graham, C., Arnold, H., Hartfield, K., & Schwenk, K. (2006). Relation of emotional reactivity and regulation to childhood stuttering. *Journal of Communication Disorders, 39*(6), 402–423. https://doi.org/10.1016/j.jcomdis.2005.12.004

Kernick, D. (2003). Introduction to health economics for the medical practitioner. *Postgraduate Medical Journal, 79,* 147–150. https://doi.org/10.1136/pmj.79.929.147

Kraft, S. J., Ambrose, N., & Chon, H. (2014). Temperament and environmental contributions to stuttering severity in children: The role of effortful control. *Seminars in Speech and Language, 35*(2), 80–94. https://doi.org/10.1055/s-0034-1371753

Kraft, S. J., Beilby, J., & Lowther, E. (2019). The role of effortful control in stuttering severity in children: Replication study. *American Journal of Speech-Language Pathology, 28*(1), 14–28. https://doi.org/10.1044/2018_AJSLP-17-0097

Langevin, M., Packman, A., & Onslow, M. (2010). Parent perceptions of the impact of stuttering on their preschoolers and themselves. *Journal of Communication Disorders, 43*(5), 407–423. https://doi.org/10.1016/j.jcomdis.2010.05.003

Markie-Dadds, C., Turner, K., & Sanders, M. (2015). *Every parent's group workbook: Triple P positive parenting solutions* (3rd ed.). Triple P International.

Mazzucchelli, T., & Sanders, M. (2018). Children with externalizing behavior problems. In M. Sanders & T. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system* (pp. 85–98). Oxford University Press.

Mazzucchelli, T. G. (2018). Social learning influences: Modelling, instructions, consequences. In M. Sanders & A. Morawska (Eds.), *Handbook of parenting and child development across the lifespan* (pp. 67–96). Springer.

Mazzucchelli, T. G., & Sanders, M. R. (2010). Facilitating practitioner flexibility within an empirically supported intervention: Lessons from a system of parenting support. *Clinical Psychology Science and Practice, 17*(3), 238–252. https://doi.org/10.1111/j.1468-2850.2010.01215.x

McAllister, J. (2016). Behavioural, emotional and social development of children who stutter. *Journal of Fluency Disorders, 50,* 23–32. https://doi.org/10.1016/j.jfludis.2016.09.003

McCulloch, J., Swift, M., & Wagnitz, B. (2017). Case file audit of Lidcombe program outcomes in a student-led stuttering clinic. *International Journal of Speech-Language Pathology, 19*(2), 165–173. https://doi.org/10.3109/17549507.2016.1159336

McGoey, K., DuPaul, G., Haley, E., & Shelton, T. (2007). Parent and teacher ratings of attention-deficit/hyperactivity disorder in preschool: The ADHD Rating Scale-IV Preschool Version. *Journal of Psychopathology and Behavioral Assessment, 29,* 269–276. https://doi.org/10.1007/s10862-007-9048-y

Millard, S., Nicholas, A., & Cook, F. (2008). Is parent–child interaction therapy effective in reducing stuttering? *Journal of Speech, Language, and Hearing Research, 51*(3), 636–650. https://doi.org/10.1044/1092-4388(2008/046)

Millard, S. K., & Davis, S. (2016). The Palin Parent Rating Scales: Parents' perspectives of childhood stuttering and its impact. *Journal of Speech, Language, and Hearing Research, 59,* 950–963. https://doi.org/10.1044/2016_JSLHR-S-14-0137

Millard, S. K., Zebrowski, P., & Kelman, E. (2018). Palin Parent–Child Interaction Therapy: The bigger picture. *American Journal of Speech-Language Pathology, 27,* 1211–1223. https://doi.org/10.1044/2018_AJSLP-ODC11-17-0199

Miller, A., Gouley, K., Seifer, R., Dickstein, S., & Shields, A. (2004). Emotions and behavior in the Head Start classroom: Associations among observed dysregulation, social competence, and preschool adjustment. *Early Education and Development, 15*(2), 147–165. https://doi.org/10.1207/s15566935eed1502_2

Moffitt, T., Arseneault, L., Belsky, D., Dickson, N., Hancox, R. J., Harrington, H., Houts, R., Poulton, R., Roberts, B. W., Ross, S., Sears, M. R., Thompson, W. M., & Caspi, A. (2011). Gradient of childhood self-control predicts health, wealth, and public safety. *Proceedings of the National Academy of Sciences of*

Ds

the United States of America, 108(7), 2693–2698. https://doi.org/10.1073/pnas.1010076108

Neece, C., Green, S., & Baker, B. (2012). Parenting stress and child behavior problems: A transactional relationship across time. *American Journal on Intellectual and Developmental Disabilities, 117*(1), 48–66. https://doi.org/10.1352/1944-7558-117.1.48

Norris, L. (2016). *Self-regulation strategies for students with disruptive behavior disorders* [Master's thesis, St. Cloud State University]. theRepository at St. Cloud State: Culminating Projects in Special Education.

NSCAW. (2009). *Inhibitory control abilities among young children in the child welfare system*. U.S. Department of Health & Human Services, Administration for Children & Families: National Survey of Child and Adolescent Well-Being.

Ntourou, K., Anderson, J., & Wagovich, S. (2018). Executive function and childhood stuttering: Parent ratings and evidence from a behavioral task. *Journal of Fluency Disorders, 56,* 18–32. https://doi.org/10.1016/j.jfludis.2017.12.001

Oberklaid, F. (2014). Prevention and early detection in young children: Challenges for policy and practice. *The Medical Journal of Australia, 201*(7), 369–370. https://doi.org/10.5694/mja14.01200

Onslow, M., & O'Brian, S. (2013). Management of childhood stuttering. *Journal of Paediatrics and Child Health, 49*(2), 112–115. https://doi.org/10.1111/jpc.12034

Petrovic, P., & Castellanos, F. (2016). Top-down regulation from ADHD to emotional instability. *Frontiers in Behavioral Neuroscience, 10,* 70. https://doi.org/10.3389/fnbeh.2016.00070

Posner, M. I., & Rothbart, M. K. (2009). Toward a physical basis of attention and self-regulation. *Physics of Life Review, 6*(2), 103–120. https://doi.org/10.1016/j.plrev.2009.02.001

Power, T. J, Costigan, T. E, Leff, S. S., Eiraldi, R. B., & Landau, S. (2001). Assessing ADHD across settings: Contributions of behavioral assessment to categorical decision making. *Journal of Clinical Child & Adolescent Psychology, 30*(3), 339–412. https://doi.org/10.1207/S15374424JCCP3003_11

Reid, R., Trout, A., & Schartz, M. (2005). Self-regulation interventions for children with attention deficit/hyperactivity disorder. *Exceptional Children, 71,* 361–377.

Reilly, S., Onslow, M., Packman, A., Cini, E., Conway, L., Ukoumunne, O., Bavin, E. L., Prior, M., Eadie, P., Block, S., & Wake, M. (2013). Natural history of stuttering to 4 years of age: A prospective community-based study. *Pediatrics, 132*(3), 460–467. https://doi.org/10.1542/peds.2012-3067

Riley, G., & Riley, J. (2000). A revised component model for diagnosing and treating children who stutter. *Contemporary Issues in Communication Science and Disorders, 27,* 188–199. https://doi.org/10.1044/cicsd_27_F_188

Rothbart, K., Ahadi, S., Hershey, K., & Fisher, P. (2001). Investigations of temperament at three to seven years: The Children's Behavior Questionnaire. *Child Development, 72*(5), 1394–1408. https://doi.org/10.1111/1467-8624.00355

Rothbart, K., Sheese, B., Rueda, R., & Posner, M. (2011). Developing mechanisms of self-regulation in early life. *Emotion Review, 3*(2), 207–213. https://doi.org/10.1177/1754073910387943

Sameroff, A. (2010). A unified theory of development: A dialectic integration of nature and nurture. *Child Development, 81*(1), 6–22. https://doi.org/10.1111/j.1467-8624.2009.01378.x

Sanchez, M., Lavigne, R., Romero, J., & Elosegui, E. (2019). Emotion regulation in participants diagnosed with attention deficit hyperactivity disorder, before and after an emotion regulation intervention. *Frontiers in Psychology, 10,* 1092. https://doi.org/10.3389/fpsyg.2019.01092

Sanders, M., Kirby, J., Tellegen, C., & Day, J. (2014). The Triple P-Positive Parenting Program: A systematic review and meta-analysis of a multi-level system of parenting support. *Clinical Psychology Review, 34*(4), 337–357. https://doi.org/10.1016/j.cpr.2014.04.003

Sanders, M. R., & Mazzucchelli, T. G. (2018). *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system*. Oxford University Press.

Sanders, M. R., Morawska, A., Haslam, D. M., Filus, A., & Fletcher, R. (2014). Parenting and Family Adjustment Scales (PAFAS): Validation of a brief parent-report measure for use in assessment of parenting skills and family relationships. *Child Psychiatry & Human Development, 45*(3), 255–272. https://doi.org/10.1007/s10578-013-0397-3

Shaw, D. S., Owens, E. B., Giovannelli, J., & Winslow, E. (2001). Infant and toddler pathways leading to early externalising disorders. *Journal of the American Academy of Child & Adolescent Psychiatry, 40*(1), 36–43. https://doi.org/10.1097/00004583-200101000-00014

Smith, A., & Weber, C. (2016). Childhood stuttering: Where are we and where are we going? *Seminars in Speech and Language, 37*(4), 291–297. https://doi.org/10.1055/s-0036-1587703

Smith, A., & Weber, C. (2017). How stuttering develops: The multifactorial dynamic systems pathways theory. *Journal of Speech, Language, and Hearing Research, 60,* 2483–2505. https://doi.org/10.1044/2017_JSLHR-S-16-0343

Smith-Donald, R., Raver, C., Hayes, T., & Richardson, B. (2007). Preliminary construct and concurrent validity of the Preschool Self-regulation Assessment (PSRA) for field-based research. *Early Childhood Research Quarterly, 22*(2), 173–187. https://doi.org/10.1016/j.ecresq.2007.01.002

Spencer, T., Faraone, S., Surman, C., Petty, C., Clarke, A., Batchelder, H., Wozniak, J., & Biederman, J. (2011). Toward defining deficient emotional self-regulation in children with attention-deficit/hyperactivity disorder using the Child Behavior Checklist: A controlled study. *Posgraduate Medical Journal, 123*(5), 50–59. https://doi.org/10.3810/pgm.2011.09.2459

Starkweather, W. C. (1987). *Fluency and stuttering*. Prentice-Hall.

Starkweather, W. C. (2002). The epigenesis of stuttering. *Journal of Fluency Disorders, 27*(4), 269–287. https://doi.org/10.1016/S0094-730X(02)00144-4

Starkweather, W. C., Ridener Gottwald, S., & Halfond, M. (1997). *Stuttering prevention: A manual for parents*. https://www.mnsu.edu/comdis/kuster/Parents/starkweather.html

Tominey, S., & McClelland, M. (2011). Red light, purple light: Findings from a randomized trial using circle time games to improve behavioral self-regulation in preschool. *Early Education and Development, 22*(3), 489–519. https://doi.org/10.1080/10409289.2011.574258

Trajkovski, N., O'Brian, S., Onslow, M., Packman, A., Lowe, R., Menzies, R., Jones, M., & Reilly, S. (2019). A three-arm randomized controlled trial of Lidcombe Program and Westmead Program for early stuttering interventions. *Journal of Fluency Disorders, 61,* 105708. https://doi.org/10.1016/j.jfludis.2019.105708

Tsukayama, E., Toomey, S. L., Faith, M. S., & Duckworth, A. L. (2010). Self-control as a protective factor against overweight status in the transition from childhood to adolescence. *Archives of Pediatrics & Adolescent Medicine, 164*(7), 631–635. https://doi.org/10.1001/archpediatrics.2010.97

Ullebø, A., Posserud, M.-B., Heiervang, E., Gillberg, C., & Obel, C. (2011). Screening for the attention deficit hyperactivity disorder phenotype using the strength and difficulties questionnaire. *European Child & Adolescent Psychiatry, 20*(9), 451–458. https://doi.org/10.1007/s00787-011-0198-9

Vanryckeghem, M., Brutten, G. J., & Hernandez, L. M. (2005). A comparative investigation of the speech-associated attitude of

preschool and kindergarten children who do and do not stutter. *Journal of Fluency Disorders, 30*(4), 307–318. https://doi.org/10.1016/j.jfludis.2005.09.003

Walden, T., Frankel, C., Buhr, A., Johnson, K., Conture, E., & Karrass, J. (2012). Dual diathesis–stressor model of emotional and linguistic contributions to developmental stuttering. *Journal of Abnormal Child Psychology, 40*(4), 633–644. https://doi.org/10.1007/s10802-011-9581-8

Webster-Stratton, C., Reid, J., & Beauchaine, T. (2013). One-year follow-up of combined parent and child intervention for young children with ADHD. *Journal of Clinical Child & Adolescent Psychology, 42*(2), 251–261. https://doi.org/10.1080/15374416.2012.723263

Wiig, E., Secord, W., & Semel, E. (2006). *Clinical Evaluation of Language Fundamentals Preschool–Second Edition, Australian and New Zealand standardised edition (CELF-P2 Australian and New Zealand)*. Pearson.

Yaruss, J. S., Coleman, C., & Hammer, D. (2006). Treating preschool children who stutter: Description and preliminary evaluation of a family-focused treatment approach. *Language, Speech, and Hearing Services in Schools, 37,* 118–136. https://doi.org/10.1044/0161-1461(2006/014)

Link between Paper 2 and Paper 3

In Paper 2 (presented in Chapter 4 of this thesis), an integrated stuttering and self-regulation program was trialled for CWS with elevated ADHD symptoms. CWS with elevated symptoms who received the novel integrated program showed significantly lower stuttering severity at three months follow-up, and also required fewer stuttering treatment sessions to achieve this more desirable treatment outcome compared to another group of CWS with elevated symptoms who received stuttering treatment only. These findings were strengthened by the large improvements in parenting practices and child self-regulation measures that occurred following the integrated program.

It is important that interventions are not only efficacious, but they are also socially valid and acceptable to those who receive them. Therefore, qualitative reflections were gathered from parents who implemented this integrated program with their children. Authors were particularly interested in capturing the perceived acceptability of this novel program for this population, as well as parent reflections on the possible interrelated improvements in stuttering and self-regulation, and how this may have manifested in real life scenarios. The data pertaining to these qualitative interview reflections are presented in Paper 3 (Chapter 5).
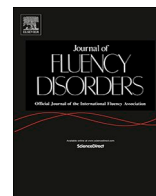
Chapter 5

Paper 3: Parent perceptions of an integrated stuttering treatment and behavioral self-regulation program for early developmental stuttering

Parent perceptions of an integrated stuttering treatment and behavioral self-regulation program for early developmental stuttering

Kerianne Druker[a], Trevor Mazzucchelli[b], Neville Hennessey[a], Janet Beilby[a,*]

[a] School of Occupational Therapy, Social Work and Speech Pathology, Curtin University, Western Australia, Australia
[b] School of Psychology, Curtin University, Western Australia, Australia

A B S T R A C T

Purpose: Recent research has identified approximately half of children who stutter present with self-regulation challenges. These manifest in elevated inattentive and/or impulsive behaviours, aligned with attention deficit hyperactivity disorder (ADHD) symptoms. These symptoms have been found to influence the child's responsiveness to their stuttering treatment, and may exacerbate the psychosocial consequences of stuttering for them and their families. Early stuttering intervention identifies parents as key agents of change in the management of their children's stuttering. This study sought feedback from parents regarding their experiences with an integrated stuttering treatment and behavioral self-regulation program for early developmental stuttering, addressing the child's self-regulation challenges.
Method: Eight parents of children who stutter who had co-occurring self-regulation challenges completed the integrated program. This incorporated the Triple P–Positive Parenting Program adapted for the developmental stuttering population, and the Curtin University Stuttering Program (CUSP). Semi-structured qualitative interviews were conducted to capture parents' reflections on, and experiences with, the integrated program.
Results: Thematic analysis identified several major themes regarding the parents' experiences with the integrated program: emotional impact on parents, child self-regulation, link between stuttering and behaviour, parent self-regulation, impact on family dynamics, and overall positive perceptions of the integrated program. All of the parents indicated they would recommend the program to future parents of children who stutter.
Conclusion: This study provides insights into parents' perceptions regarding an integrated intervention approach for early stuttering and behavior management. It also indicates how adopting a holistic approach to stuttering intervention is positive and has social validity.

Introduction

The multifactorial nature of developmental stuttering disorders has been highlighted in contemporary research with the examination of a range of factors that contribute to the onset and prognosis of this complex disorder (Smith & Weber, 2016, 2017). One factor that has received recent, significant attention is the temperament of the child. Some children who stutter have been found to

exhibit increased emotional reactivity and poorer self-regulation skills compared to their fluent peers (Conture, Kelly, & Walden, 2013; Eggers, De Nil, & Van Den Bergh, 2013; Eggers, De Nil, & Van den Bergh, 2013; Eggers, De Nil, & Van Den Bergh, 2012; Jones, Choi, Conture, & Walden, 2014; Karrass et al., 2006). Furthermore, it has been demonstrated that poorer self-regulation skills in these children, correlates with more severe stuttered speech (Kraft, Ambrose, & Chon, 2014; Kraft, Beilby, & Lowther, 2018). Research has found that half the children who stutter presenting to clinics for stuttering treatment have concomitant self-regulation challenges (Druker, Hennessey, Mazzucchelli, & Beilby, 2019), manifesting in elevated (subclinical) levels of attention deficit hyperactivity disorder (ADHD) symptoms (Donaher & Richels, 2012).

Emotional dysregulation is a core deficit in ADHD (Barkley, 2013). Therefore, although these children who stutter do not necessarily meet criteria for an ADHD diagnosis, such elevated ADHD symptoms have been found to predict reduced stuttering treatment responsiveness (Druker, Hennessey, Hennessey et al., 2019; Riley & Riley, 2000). Furthermore, Felsenfeld, Van Beljsterveldt, and Boomsma (2010)) have suggested the possibility that "unstable fluency is part of a broader spectrum of regulatory problems" (p. 1162) and have highlighted the need for "clinicians to adapt their therapeutic strategies to support children with both stuttering and attentional disorders" (p. 1162). The importance of addressing self-regulation skills in speech, language and communication disorders, to best support the child's engagement and success in therapy, has been emphasized in recent translational research (Binns, Hutchison, & Oram Cardy, 2019).

Children with diagnosed ADHD are known to have increased behavioral and emotional problems (Barkley, 2013). Parents of children with ADHD have been shown to experience higher levels of parenting stress, increased martial conflict, greater incidence of maternal depression, and inconsistent expectations across siblings (King, Alexander, & Seabi, 2016; Peters & Jackson, 2008; van Steijn, Oerlemans, van Aken, Buitelaar, & Rommelse, 2014). Mothers of children with ADHD have been found to be more negative and directive, less rewarding of desirable child behaviors, as well as less responsive to their children's questions (Danforth, Barkley, & Stokes, 1991; Ellis & Nigg, 2009). Furthermore, parent-child interactions have been found to be more negative and hostile than in comparable families (DuPaul, McGoey, Eckert, & VanBrakel, 2001; Neece, Green, & Baker, 2012). These difficulties are important to consider, as negative and inconsistent maternal discipline, maternal mental health problems and marital conflict are a consistent set of risk factors found to be associated with the continuity of preschool behavior problems (Campbell, 2000).

Psychosocial impact

Poor self-regulation skills, manifesting in elevated ADHD symptoms (Barkley, 2013), in the pre-school ages have been found to predict poor long-term academic, social, health and employment outcomes (Moffitt et al., 2011; Tsukayama, Toomey, Faith, & Duckworth, 2010). Therefore, beyond the impact of poor self-regulation skills on stuttering outcomes, a range of psychosocial consequences found to be experienced by families of children with ADHD may also be experienced by the families of these young children who stutter. This may compound the already well-documented negative psychosocial impact experienced by children and their families as a consequence of living with a stuttering disorder (Beilby, 2014; Langevin, Packman, & Onslow, 2010; Millard & Davis, 2016; Plexico & Burrus, 2012).

Self-regulation and the Demands and Capacities Model

Children with poor self-regulation skills are less able to successfully regulate their emotions, which in turn may lead to increased emotional reactivity in response to environmental changes (Eisenberg & Sulik, 2012). Although only speculative in nature, improved self-regulation skills, likely to result in less impulsive and volatile behavior, can be considered in light of the Demands and Capacities Model (DCM; Starkweather, 1987). The DCM posits that stuttering onsets and is maintained when the capacity for fluent speech production is outweighed by the child's excessive internal demands or the external environmental demands they experience. According to this model, decreased impulsivity and emotional volatility will reduce excessive resources which can then be allocated to the regulation of emotional and motoric pressures. This ultimately frees up additional resources for fluent speech production in the child. Addressing self-regulation to support fluency is further supported by Eggers, De Nil, Van Den Bergh et al. (2013) who suggest that improvements in impulse control are likely to translate into improved emotional regulation, "decreasing the emotional arousal, and thus reducing its possible impact on the exacerbation of stuttering symptoms" (p. 9).

Parent role in therapeutic process

The role that parents play in the treatment of developmental stuttering disorders, whether direct or indirect, is well documented in the literature (De Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015; Druker, Hennessey, Hennessey et al., 2019; Druker, Mazzucchelli, & Beilby, 2019; Franken & Putker-de Bruijn, 2007; Millard, Nicholas, & Cook, 2008; Onslow & O'Brian, 2013; Yaruss, Coleman, & Hammer, 2006). In addition, the negative psychosocial consequences experienced by parents as a consequence of having a child who stutters is also recognized strongly (Langevin et al., 2010; Millard & Davis, 2016; Plexico & Burrus, 2012). Given the significant role that parents play in treatment, as well as the psychosocial effects they experience, it is important to provide parents with support to address all aspects of the stuttering disorder (Yaruss & Quesal, 2004), including the concomitant self-regulation deficits or elevated ADHD symptoms that co-occur in a large proportion of this childhood population (Druker, Hennessey, Hennessey et al., 2019). Parents play a significant role in the development of self-regulatory capacity in children (Deater-Deckard, 2014; Ullsperger, Nigg, & Nikolas, 2016), and may therefore, with guidance, be able to moderate the negative long-term consequences of self-regulation challenges present in preschool children (Moffitt et al., 2011). One previous study has attempted to concurrently

address fluency and self-regulation skills in children who stutter through an integrated stuttering treatment and resilience training program (Druker, Mazzucchelli et al., 2019). Findings yielded positive outcomes in terms of enhanced psychosocial wellbeing for children who stutter and their parents (Druker, Mazzucchelli et al., 2019).

The Triple P Program

Evidence-based parenting interventions that aim to increase positive parent-child interactions, while reducing inconsistent and conflictual parenting practices, are effective in treating a range of child behavior disorders (Eyberg, 1988; Patterson, 2005; Sanders, 2003; Webster-Stratton, 1998). Several studies examining the effectiveness of parent training in families of children with ADHD, including the Triple P–Positive Parenting Program (Hoath & Sanders, 2002) have demonstrated significant overall improvements in child compliance, reduction in non-compliance, and improvements in parenting practices (Barkley, 2013; Sonuga-Barke, Daley, Thompson, Laver-Bradbury, & Weeks, 2001).

The Triple P–Positive Parenting Program (Sanders & Mazzucchelli, 2018) has demonstrated benefits in supporting parents to encourage emotional-regulation, independence, problem solving and communication skills in their children by implementing consistent consequences for misbehavior and rewards for compliance (Bor, Sanders, & Markie-Dadds, 2002). It also promotes the development of positive parent-child interactions with the overarching purpose of promoting self-regulation in both parents and children (Sanders & Mazzucchelli, 2013). Parent self-regulation refers to a parent's capacity to monitor, judge and change their own behavior, essential for the development and maintenance of positive and nurturing parenting practices, as well as to identify and accept problems in children's development (Sanders & Mazzucchelli, 2013). Furthermore, the program has yielded improvements in communication between parents, reduced levels of parenting stress, and increased overall parenting confidence (Sanders, Kirby, Tellegen, & Day, 2014).

The present study

Extending the original pilot study by Druker, Mazzucchelli et al. (2019), a randomized controlled trial evaluating the efficacy of an integrated stuttering and behavioral self-regulation program has been conducted (Druker, Hennessey, Mazzucchelli, & Beilby, 2019). The stuttering component of the integrated program, being the Curtin University Stuttering Program (CUSP), comprised a combination of direct and indirect treatment principles (Druker, Hennessey et al., 2019, 2019b; Druker, Mazzucchelli et al., 2019; Shafiei, Faramarzi, Abedi, Dehqan, & Scherer, 2019). Triple P was delivered concurrently, as the self-regulation component of the program, with modified examples embedded throughout and an additional tip sheet illustrating how the content was to be applied relevantly for children who stutter (Druker, Hennessey et al., 2019). The Triple P component was delivered individually with parents using telehealth delivery, and stuttering treatment sessions were administered at university, private or community-based speech pathology clinics. Pre-, post,- and follow up quantitative data pertaining to stuttered speech severity, parenting practices, and child behavior, has been collected (Druker, Hennessey et al., 2019).

Parenting practices and coping skills improved for all parents in the current study (Druker, Hennessey et al., 2019). In addition, all children of parents interviewed obtained fluency within normal limits, and experienced significant improvements in behavior and self-regulation skills over the course of the integrated program (Druker, Hennessey et al., 2019).

The current study aims to capture parents' reflections of this novel integrated program, to determine the perceived acceptability and possible benefits of the integrated program for young children who stutter and their families. Patient centered care is an approach to healthcare that emphasizes partnerships between consumers and health professionals, and has emerged as a primary approach to health care delivery (Delaney, 2018). This method recognizes the input that health care consumers can have on enhancing healthcare service delivery, promoting increased patient satisfaction (Delaney, 2018). Consumer perceptions have also been shown to affect uptake, engagement, and subsequent effectiveness of an intervention (Mazzucchelli, Studman, Whittingham, & Sofronoff, 2010). As such, the qualitative nature of the present study aims to explore more fully the parents' experiences and perceptions of this client-centered approach to early stuttering treatment. It may also provide the opportunity to gather consumer information that might otherwise be lost or underestimated, potentially informing enhanced future implementation of this integrated program (Connery, McCurtin, & Robinson, 2019).

Method

Participants

Eight parent participants were recruited by convenience sampling. Inclusion criteria were: (a) primary caregiver, (b) English as a first language, (c) their child having a diagnosis of developmental stuttering disorder, (d) their child presenting with a total score at or above the 80th percentile for his/her age and sex on the ADHD Rating Scale, as well as a rating of 2 (often) or greater on any two items related to inattention or any two items related to hyperactivity/impulsivity (Druker, Hennessey et al., 2019, 2019c), (e) participant having no previous or current professional support for parenting practices or childhood behavior problems. All participants in the present study were female, with an age range of 26 to 38 years (see Table 1 for additional participant information). These participants were drawn from a larger sample (n = 20) in an associated quasi-experimental clinical trial (Druker et al., 2019a). Although all eight participants were interviewed, the final two interviews did not result in identification of any new themes, therefore saturation was achieved (Guest, Bunce, & Johnson, 2006).

Table 1
Participant demographic characteristics.

P	Sex	Relationship to child	Ethnicity	Highest level of education completed	Hours spent in paid employment 1 = none 2 = < 20 3 = 20-30 4 = 30-40 5 = > 40	Able to meet household expenses	After expenses can afford 1 = not much 2 = some things 3 = most things	Attendance in sessions M = mother F = father B = both	Marital status	No CWS	Age of CWS (years)	Sex of CWS	Total children
SF	F	Mother	Caucasian	Secondary school	4	Y	1	B	Separated	1	3	M	1
LD	F	Mother	Caucasian	Masters	2	Y	1	M	Married	1	3	M	3
KP	F	Mother	Caucasian	TAFE/ Certificate	2	Y	3	B	Married	1	3	M	2
LM	F	Mother	Caucasian	Baccalaureate	4	Y	2	B	Married	1	5	M	3
KH	F	Mother	Caucasian	Secondary school	1	Y	1	B	Married	2	3 4	M M	2
FG	F	Mother	Caucasian	Baccalaureate	1	Y	3	B	Married	1	3	M	2
SM	F	Mother	Caucasian	Secondary school	1	Y	1	M	Married	1	6	F	3
AM	F	Mother	Caucasian	Baccalaureate	3	Y	2	M	Married	2	4 6	M F	2

Note. P = participant. M = male. F = female. No = number of. CWS = children who stutter.

(insert Table 1 about here)

Procedure

Ethics approval was obtained for this study through the Curtin University Human Research Ethics Committee. Individual semi-structured interviews (lasting 25 minutes on average) were conducted with the parent participants. The semi-structured interview probe questions explored their experiences with the integrated program and its possible benefits and applicability to the population of children with early developmental stuttering. Questions were open-ended and participants were prompted to expand on their reflections as considered necessary. Questions relating to the perceived impact of the program on the parent and child were based on previous qualitative research evaluating Triple P Parenting Programs (Haskett, Armstrong, Neal, & Aldianto, 2018; Houlding, Schmidt, Stern, Jamieson, & Borg, 2012). See Appendix A for questions used to guide the interviews. An independent research assistant, blinded to the program content and study aims, conducted all interviews. All recordings were digitally recorded and transcribed verbatim.

Data analysis

The research team comprised the first author who is a final year doctoral student and certified practicing speech pathologist, and three senior researchers. Data were analyzed according to the protocol described in Braun and Clarke (2006), involving six recursive phases of data analysis (see Table 2). QSR's (2018) NVivo 12 qualitative data analysis software was used to assist in organization and analysis of the data. After initial themes were stipulated by the first author, major themes and subthemes were agreed upon by the final author. In addition, an external research assistant independently coded two transcripts selected at random using the proposed coding framework provided by the first author, and a high degree of inter-rater reliability rating was reached (93% agreement) (Barbour, 2001). The final two interviews did not result in identification of any new themes, therefore saturation was deemed as achieved (Guest et al., 2006).

Table 2
Braun and Clarke (2006) phases of qualitative research and associated activities

	Phase	Activity
1	Familiarization	Transcripts were read twice before coding commenced
2	Generation of initial codes	Significant participant statements throughout each manuscript were highlighted and assigned an initial code
3	Searching for themes	Codes were organized into potential themes
4	Reviewing themes	Themes were refined and a thematic map of the data set was developed
5	Defining and naming themes	Major themes were re-examined and relevant sub-themes were identified
6	Collation of the report	Powerful extract examples were selected, and analysis was related back to the research question and previous literature

(insert Table 2 about here)

Results

In total, 6 main themes and 29 subthemes emerged, summarized in Table 3. The number of participants that endorsed each theme, the number of occasions the theme was referenced throughout the complete data set, and an exemplar participant statement is provided in Table 3. Selected quotations from parent interviews in the table and throughout the results section address the integrated nature of the program, and most reflections encompass combined aspects of the stuttering treatment and the Triple P program component.

Table 3
Summary of major and sub-themes identified in thematic analysis

Major Theme	Sub-Theme	Endorsement of Theme (n)	References (n)	Exemplar Quote and Participant Identifier
Emotional impact on parents		8	156	
	Experiencing negative emotional reactions	8	113	"… with my frustration level with my oldest, I was at breaking point some days where I couldn't even cope. I had to separate myself from him… my stress levels were just at its peak." - KH
	Holding concern for their child's future	7	18	"I was very stressed. I would cry a lot and worry all the time about how [my son]'s behavior and his stuttering was going to affect his school, learning and friendships, and any relationship he has going forward." - KH
	Parent emotions affecting their child	4	5	"Last week, it was just so rushed and I just lost it a bit. I didn't slow down, I was stressed… it just went from bad to worse. We were both just so worked up, I see it now so clearly." - SF
	New-found emotional stability	5	20	I don't get as upset, I don't get as angry and I just kind of stay at the same level, and it makes my child levelled out too. - LD
Child self-regulation		8	116	
	Shift in child self-regulation	8	52	"He really is like a different child.. he listens better, he lashes out less, we do argue less with each other, and when we ask him to stop and slow down he really tries." – KH
	Labelling emotions and stutter	4	8	"… when he developed the language that accompanied his emotions, he could self-regulate so much better and that was a huge turning point for him" - FG
	Pre-planning outings or challenging situations	6	14	"I now always try to make a point of spending time talking about the rules before an outing that is usually very stressful, and then he knows what's expected of him and he knows what he has to do to get his reward."- LD
	Safety within boundaries	6	31	"It stops either the tantrum or the shouting or the storming off to her room… because she realises earlier that the ball's back in my court and if I can do what mummy's asked me to do, it's not going to advance to the next level." - SM
	Focus on positives	4	11	"actively seeking ways to let [him] know that he's doing a good job, so giving him positive reinforcement as we go along which really, really works for [my child].
Link between stuttering and behavior		8	101	
	Stutter elicits negative feelings	8	21	"He was very frustrated because he had a lot to say and it wasn't coming out the way he imagined it… that would affect his mood and demeanour and behavior so it was all a bit of a spiral." - FG
	Emotions impact fluency	8	36	"If he was allowed to spiral into a tantrum, then he would start stuttering… so it was like whatever head space you get into with those intense emotions that make a child tantrum would make him stutter." - FG
	Bidirectional relationship between stuttering and behavior	8	29	"I think him being able to stop and regulate himself has helped him to talk and explain himself much better without getting frustrated and starting to stutter, or vice versa. I think it all works hand in hand and it just works well together." - KP
	Improved behavior shapes improved fluency	7	16	"It goes hand in hand – the talking and the behavior. If you don't have control over their behavior, how can you help them with their talking or how can they help themselves with their talking if they just do whatever they want to do." - SF
Parent self-regulation		7	169	
	Lack of capacity and knowledge prior to intervention	7	38	"I didn't want to be a bad mum.. I just didn't know how to manage him, he was just out of control and he walked all over me." – SF
	Parent in control	7	38	"I feel like I've become a lot more in control of his behaviour as I now realise that I can affect his behaviour by the way I manage it." - LM
	Consistency in parenting practices	5	21	"I think they feel that mummy means business now… before I gave in a lot more." – SM

Table 3 (*continued*)

Major Theme	Sub-Theme	Endorsement of Theme (n)	References (n)	Exemplar Quote and Participant Identifier
	Confidence in parenting ability and ability to manage stuttering	5	16	"I know now how to deal with situations, and it makes me more confident and more relaxed around him… I feel like I'm the same as his dad now, I don't have to stand back." - SF
	Realistic expectations	7	20	"I was just at my wits end for the day, and normally I would be so angry at him I wouldn't even be able to look at him, but I remembered all the stuff I needed to do… so I didn't hold the anger. I just let it go and carried on with my day." - KH
	Experiencing growth and change	7	38	"Before I started this program, I was feeling very unnerved and very worried. So it's not only helped me manage the kids, its actually helped me manage myself and have a bit of direction to know that I'm doing the right thing to push forward." - KP
Family dynamics		6	66	
	As a whole family	5	8	"We've been a lot more calm in the house, so there's been a bit more happiness generally." - LM
	Parents working together	6	13	"This program has made [my husband] realise that he needs to be more on board… so it's definitely a first step and I think he finally recognises that it won't work if its left all up to me." - KP
	Enhanced parent-child relationships and interactions	5	36	"Even just the last few weeks of giving him more love, cuddling him more, being more around him and enjoying being around him, it's changed our relationship dramatically and in turn it's changed his behavior." - KH
	Flow-on effect to siblings	4	9	"It's been nice because they're able to work together to achieve things that they both want, so it's sort of provided a bit more unity between the two boys." - LM
Positive perceptions of integrated program	Positive perceptions of integrated program	6	86	
	Early intervention	4	5	"As soon as we started treatment, I felt more confident about it so I didn't have a lot of time to feel worried because we got him in pretty quickly." - LD
	Therapist support	4	7	"It's great to have an actual professional's opinion… and sometimes it can be so simple and so life-changing." - KH
	Ease of delivery	2	4	"The fact that [the therapist] could deliver it to me over Skype at a time convenient to me… that's all the difference for me, that enabled me to participate in it." - SM
	Naturalistic approach to intervention	5	17	"It's made it so easy that I can fit it into my life." - KP
	Surprise at process	5	27	"It was really just a couple of days of being extra strict with [my son], and really following through, did I see such a massive improvement in his response to me and his listening." - AM
	Program seen as a life-changing experience	6	27	"It has changed, changed everything for me because I was a wreck like I was at my absolute wits end… it has been life changing for us like a big, big difference." - KH

(insert Table 3 about here)

Theme: Emotional impact on parents

As parents shared their stories, they all reported the significant emotional impact experienced as a result of having child who stutters with self-regulation challenges. This involved the negative emotional reactions experienced and the concern for their children's future. Following their participation in the program, the majority of parents discussed the realization that their own emotions affect their children but they reported new-found emotional stability.

Experiencing negative emotional reactions

Every participant reflected on their negative emotional reactions in response to their child's stuttering and/or self-regulation challenges. In response to their children's stuttering, parents reported feelings of anxiety, guilt, frustration, sadness, stress, lack of control, and initial uncertainty regarding how to manage the disorder. In response to their children's behavior problems, parents reported all of the aforementioned feelings associated with having a child who stutters, in addition to feelings of anger, despair, shame, exhaustion and difficulty coping were reported. The following quotes exemplify this subtheme:

> KP: *I think maybe I could have done something, you know, all those emotions you get as a parent…the guilt.*

> KH: *I was crying all the time I just, he was driving me crazy…I [didn't] even know what to do.*

Holding concern for their child's future

Seven out of eight parents discussed the concerns they held for their children's future, in terms of developing friendships, learning, and possible future relationships, as demonstrated below:

> KH: *I was thinking how is he going to go at school like playing with other kids, like he couldn't get the words out so they will eventually be like you're too hard to play with'cos you don't talk.*

Parent emotions affecting their child

Half the parents reflected on the realization of the impact of their own emotional state on their children's mood and volatility, as well as their children's fluency. Below is one reflection:

> SF: *Just from being calm and quiet and explaining things again he's back to normal again…his stuttering went up to about a 7 or 8 - it was every word of every sentence and it was stuck like he couldn't get the words out and now, yesterday, he's a three. So, big difference.*

New-found emotional stability

Five parents shared their journey to becoming more successful in terms of the way they managed their own emotions, reflected below:

> KP: *It makes me stop and think about how can I problem-solve a tantrum or a misbehavior. At least it makes me stop and think about it, so I'm not feeling nearly as guilty as a parent.*

Theme: Child self-regulation

One of the most common themes reflected by parents in the interviews was the shift in their children's self-regulation skills over the course of the integrated program. This was supported by the children labelling their emotions and their stuttered speech, parents taking time to pre-plan outings, providing safety within boundaries, and focusing on positive behaviors.

Shift in child self-regulation

The shift in child self-regulation was reported in all of the interviews. Parents spoke about their children's difficulties with self-regulation before the program and provided the following examples: constant meltdowns and emotional outbursts, impulsivity, non-compliance, excessive talking, aggression, snatching, extreme anger and frustration. The shift in child self-regulation was highlighted as parents reflected on their children after the program: listening better, taking ownership of their behavior through apologizing, being less needy, having less aggressive outbursts and tantrums, not getting into trouble as frequently, respecting others' personal space, improving in their ability to delay gratification, slowing down, thinking about the consequences of their behavior, stopping an undesirable behavior after a warning, and calming themselves down:

> LD: *…he is getting much better at calming himself down. So, if he gets to go to quiet time with his Lovey, which is his blanket, it kind of resets him, and that can also help with his fluency.*

Labelling emotions and stutter

Half the parents found that giving the child the language to identify their emotions, as well as their stutters or "bumpy words",

was crucial in supporting the child's awareness and positive understanding. Emotional labelling by the child helped parents to guide them more appropriately and calmly through volatile outbursts. This subtheme is illustrated by the following quote:

FG: *…giving him the language to help him identify his feelings and where he's at would allow us to guide him appropriately…and so if he was doing something that was making him angry…instead of just losing control he would say 'I'm frustrated by that', or 'I feel angry', or 'I feel upset', and that has really helped.*

Pre-planning outings or challenging situations

The merit of pre-planning outings or challenging situations was discussed in six out of eight interviews. By having pre-planned expectations and consequences, there was less avoidance of previously high-pressure and potentially negative outings such as grocery shopping. The following quote supports this subtheme:

LM: *I think just putting in a little bit of thought into those high-pressure situations really paid off for us. So, yeah that was a good breakthrough moment for us…and if the behavior's not escalating the stutter's not going to escalate.*

Safety within boundaries

Six parents reflected on the importance of setting clear boundaries to support the development of their children's self-regulation skills. Parents reported that as a result of the lack of boundaries pre-program, children were "walking all over [them]" and often escalated the behavior problems because they "never knew what the outcome would be". Setting clear boundaries helped children to learn the link between their behavior and consequences and increased stability in the home, as "the kids [knew] where they stand" and expected the "same outcome". The following quote further illustrates this subtheme:

SM: *I think it's allowed her to realize earlier on if I do as I'm told it's not going to end up in a tantrum or it's not going to end up with mummy shouting at me…and that helps her fluency.*

LM reported an initial increase in disruptive behavior as her child "tested the boundaries" and rebelled against the newly implemented consequences, however this decreased when boundaries remained stable:

I feel like he has more of an expectation that the behavior will be managed so the more, the longer, we are putting these strategies in place, the more he expects them and he's not rebelling so much against them.

Focus on positives

Half of the parents who were interviewed reflected on the importance of focusing on the desirable aspects of their child's behavior and fluency to encourage development of their child's self-regulation skills, as reflected on below:

LD: *I do focus on [the] positive more than [the] negative now…he's more susceptible with the positive feedback and gets really excited about it.*

Theme: Link between stuttering and behavior

All of the parents provided a positive response when they were asked if they believed there was a link between their children's stuttering and their behavior and emotions in their daily lives. Parents reflected on the notion that stuttering elicits negative feelings in their children, emotions impact fluency, the existence of a bidirectional relationship between stuttering and behavior, and the experience of improved child behavior shaping improved fluency.

Stutter elicits negative feelings

All parents expressed the idea that they felt stuttering elicits negative feelings in their children, as illustrated in the following quote:

KP: *…he couldn't get his words out, he was getting so frustrated, and his behavior and tantrums were out of control.*

Emotions impact fluency

The supposition that positive and negative emotional responses exacerbate fluency was reflected on in all eight interviews, as expressed below:

LM: *…so when he's stressed out, when he's tired, when he has gotten in trouble a lot at school, I can tell because the stutter is quite profound at home.*

Bidirectional relationship between stuttering and behavior

Once again, each parent expressed their belief in a bidirectional relationship between stuttering and behavior:

AM: *I don't often know what starts off the other when he's having a bad patch? It's very difficult to know if it's the behavior then the stutter, or the stutter then the behavior. Sometimes it's very difficult to work out what's triggering what.*

Improved behavior shapes improved fluency

Seven out of eight parents spoke about the positive effects that improved behavior had directly on their children's fluency, as highlighted below:

LM: *I think this program is excellent for giving the parents strategies to manage behavior and so that it doesn't escalate and so I think that's if the behavior's not escalating the stutter's not going to escalate.*

Theme: Parent self-regulation

Parent self-regulation emerged as a major qualitative theme in the results. All of the parents reported a lack of confidence in dealing with their children's difficulties pre-program, because they felt they lacked knowledge and capacity. Over the course of the integrated program, the journey to enhanced self-regulation was achieved through parents taking control, implementing consistent parenting practices, becoming more confident and having more successful and realistic expectations of themselves and their children. Parents spoke of this overarching concept in the context of both behavior and stuttering management.

Lack of capacity and knowledge prior to program

Seven parents reflected on their lack of knowledge and understanding contributing to their lack of overall parenting confidence, all seven in relation to their child's behavior and six in relation to their child's stuttering, as reflected below:

LM: *We just felt helpless…we had no idea where to start.*

Parent in control

Seven parents felt that they gained control over their own reactions as well as their children's behavior, as expressed below:

SM: *…the kids know where they stand and they know that mummy's not going to take it…*

Consistency in parenting practices

Five out of eight parents reflected on their increased consistency in parenting practices, illustrated by the following quote:

AM: *He just learnt very quickly that if I said no, he can't do that, he actually wouldn't do that. Because he learnt that whatever I said wouldn't happen actually wouldn't happen.*

Confidence in parenting ability and ability to manage stuttering

Five parents revealed an increase in parenting confidence, in terms of managing their children's self-regulation difficulties as well as their stuttering.

SF: *…you just feel stronger and so much confident.*

Realistic expectations

Seven parents reflected on the importance of having realistic expectations on their own parenting skills, as well as their child's behavior and fluency progress.

KP: *The strategies are becoming easier, but I guess there's always going to be situations where you don't get the outcome you want. And I guess that's putting a lot on the child, so I guess expectations comes into it as well.*

Experiencing growth and change

Seven parents discussed the growth and change they experienced, in terms of how they manage their own emotions and their children's difficulties, as reflected below:

AM: *Having more knowledge and understanding has been a huge help…I now know what to do and am coping better.*

Theme: Family dynamics

Parents discussed the impact of having a child who stutters with concomitant self-regulation problems on the whole family dynamics, and how this transformed over the course of the program. Parents discussed the positive effects on the whole family, on parents working together, on enhancing parent-child relationship and interactions, as well as the flow-on benefits to siblings.

As a whole family

This subtheme emerged in five interviews, when parents were reflecting on the broader impact of their child's difficulties. Quotes below highlight the positive shift in family dynamics from pre (SF) to post (SM) program:

SF: *When the stuttering was severe it strained the family dynamics, imposed an added stress and gave doubt [as] to whether [my child] would ever normalize.*

SM: *It's made for a lot more peace and a lot more stability and, um, yeah without a doubt it's made a huge difference.*

Parents working together

Six out of eight responses in the parent interviews expressed the perceived importance of partner support in managing their children's stuttering and behavior problems, and in some cases, the transition to partners becoming increasingly more supportive. The first extract from AM's interview discusses the lack of partner support and associated burden felt when she was engaged in a previous stuttering treatment approach and was not working together with her partner.

AM: *…when I was the only one to have the half hour sessions, that's when my husband felt like I wasn't doing enough. I felt like it was all on me and I was frustrated with my husband.*

LD discussed the transition to her and her partner becoming aligned with their new approach to managing their child's behavior, as a result of the integrated program:

I would try to do one way of parenting, he might do another and so we weren't on the same page of what to do because we both probably just wanted to help him in the moment…so that has changed. [My husband's] on board now and so we're both on the same page of what to do and how to handle it rather than just having our own approach separately.

Enhanced parent-child relationships and interactions

Six parents reflected on the transition to improved parent-child relationship over the course of the program. Five parents reported strained parent-child interactions pre-program as a consequence of their child's self-regulation challenges, and one felt her child's stutter compounded this strain on their relationship.

AM: *I found it very difficult to have a connection with her, because I found her so frustrating…that definitely affected the bond at certain times.*

Flow-on effect to siblings

Half of the parents reflected on the flow-on effects to siblings following the integrated program, in terms of behavior and fluency management principles, improved sibling relationships, and more stable parent expectations for all siblings.

KH: *Because we were helping our little one out all the time after being taught what to do, the oldest one picked up on [it]…, he just followed through helpfully and naturally with what we were doing with the little one which was great.*

Theme: Positive perceptions of integrated program

Another major theme identified was the positive parent perceptions of the integrated stuttering treatment and behavioral self-regulation program. All parents answered positively, when asked if they felt the integrated program may benefit future parents of children who stutter, with one parent adding the caveat that she felt it would only be beneficial if the child also had behavior challenges. A number of factors contributed to these positive perceptions including: parents recognizing the importance of early intervention, therapist support, ease of program delivery, the naturalistic approach to intervention, surprise at the overall process and the program being seen as a life-changing experience.

Early intervention

Half the parents reflected that they felt that early intervention for stuttering as well as self-regulation challenges was optimal for treatment success, as well as for alleviating the consequent short- and long-term negative psychosocial impact on the child and other family members. The following quote illustrates the continuous parent anxiety that may have ensued in the absence of the early stuttering intervention.

FG: *I know there might be a large percentage that go through it on their own and it just naturally abates but you don't know what category your child is going to fit in, is it going to come back when they're three or four?*

Therapist support

A number of participants reflected on the importance of therapist support, in terms of providing parents with realistic expectations, offering professional advice and education, encouraging problem solving between parent dyads, as well as helping parents to feel less isolated in managing their children's difficulties. This subtheme is illustrated by the following quote:

SF: *…[the therapist] makes you realize you know you're not in it by yourself…a lot of parents go through it too.*

Ease of delivery

Three parents reflected on the ease of telehealth delivery of the Triple P component of the program being an enabling factor in their participation in the integrated program, in terms of access and resources provided.

SM: *…physically getting to the [previously offered Triple P] course in the one or two days a week in that certain time slot that they wanted was impossible for me. I just, I couldn't manage that with our lives…*

Naturalistic approach to intervention

Five parents reflected positively on the naturalistic intervention approach. They spoke about the program "making sense", being easy to fit into their lives, and employing "simple" yet highly effective principles for both stuttering and behavior management. This subtheme is illustrated by the following quote:

KH: *…there were simple things that now we've just implemented that have made huge changes…those simple changes have really changed our lives at home and um yeah we're really, really, really thankful that we were involved in it and happy with the result that we got.*

Surprise at process

Five parents expressed their surprise at the intervention process, including the children's responsiveness to program techniques and the naturalistic intervention principles:

KH: *As soon as we started speech therapy we noticed a difference straight away.*

Program seen as a life changing experience

Six out of eight participants referred to the program as being a transformative experience on 27 separate occasions. The term "life changing" was used by two parents on 7 separate occasions, informing the title of this subtheme. Example quote below:

SF: *Since I've been doing this, he's a whole different more positive, happier child.*

Discussion

The present study explored the experiences of parents of children who stutter with an integrated stuttering treatment and behavioral self-regulation program. The current study is unique in that it addresses self-regulation issues in the field of early stuttering treatments using an evidence-based parent training program. Given the key role that parents play in stuttering interventions, it is important to capture their reflections on this integrated program in terms of acceptability, and their insights regarding possible benefits for future clinical practice. Such reflections provide valuable insight into parents' experiences with the program, promoting concordance between health care providers and consumers, and subsequently maximizing future patient satisfaction, engagement and adherence to program principles (Bertakis & Azari, 2011; Delaney, 2018; Ekman et al., 2012). Major and minor qualitative themes which emerged in the study's interviews are discussed below.

Emotional impact on parents

The profound emotional impact experienced by parents in the current study is supported by previous qualitative research investigating parent experiences with children who stutter (Langevin et al., 2010; Plexico & Burrus, 2012), studies relating to parents of children with ADHD or behavior problems (Brown, McIntyre, Crnic, Baker, & Blacher, 2011; Neece et al., 2012), and more general research exploring emotional stress levels of parents who have children with developmental delay or disorder (Minnes, Perry, & Weiss, 2015). Parents of children who stutter with concomitant self-regulation challenges may experience compounded effects of either developmental challenge in isolation. Such support is important to address as parenting stress and mental health has been closely linked to a range of developmental outcomes in children, including development and maintenance of internalizing and externalizing behaviors (Gross, Shaw, & Moilanen, 2008; Neece et al., 2012). In this light, the journey to increased parent emotional stability reflected on in the study's interviews, characterized by a reported reduction in stress and less frequent negative emotional reactions, is a beneficial consideration for future integrated stuttering interventions.

Child self-regulation

Parents reflected that as their children improved in their self-regulation capacities, their stuttered speech improved as well; underscoring the importance of addressing stuttering from a holistic standpoint. A number of specific examples of improved child self-regulation skills provided by parents in the study's interviews were linked to improvements in speech fluency. For example, parents reflected that the integrated program helped their children to slow down in their speech rate at times. This proved beneficial given that increased demands on fluency, through increased time pressure, made the child more likely to stutter (Starkweather, Ridener Gottwald, & Halfond, 1997). In addition, a number of parents reflected that children were less likely to interrupt if they were having important conversations with their partners for example. This again supported better fluency as the child timed their contributions to conversations more successfully and they did not compete as much for attention (Starkweather et al., 1997). Furthermore, the importance of pre-planning activities was highlighted in the study's interviews in that this set clearer expectations and increased predictability of possibly "high-risk" or high pressure situations (Sanders & Mazzucchelli, 2018). Given that lack of structure and uncertainty may contribute to an increase in stuttering (Starkweather et al., 1997), pre-planning on the part of these parents decreased the uncertainty and unpredictability of certain scenarios, and subsequently reduced fluency destabilization.

Beyond the possible consequent improvements to speech fluency, the possible improvements in child self-regulation as reflected

on by parents, has been shown to be associated with positive long-term health and psychosocial outcomes, including academic achievement, physical and mental health, economic behavior and better interpersonal relationships (Moffitt et al., 2011; Sonney & Insel, 2016; Tsukayama et al., 2010).

Link between stuttering and behavior

Parent comments regarding the link between stuttering and behavior underpins the importance of an integrated intervention program respectful of these reflections. One parent commented directly on how stuttering elicited negative feelings in her child related to her speech. This is supported by research indicating pre-school children who stutter to have a more negative attitude towards their speech than their fluent peers (Vanryckeghem, Brutten, & Hernandez, 2005). A number of parents highlighted how stuttering elicits more general feelings of anger and frustration in their children, as they experienced difficulty expressing themselves. Parents also reflected on the notion that emotions played a key role in exacerbating their children's stuttering. This is supported by Walden et al. (2012) who found situational stressors that give rise to emotional arousal are linked to occurrences of stuttering, and concluded that "emotional arousal in itself, whether positive or negative, disrupts fluency for children who stutter" (p.11). In line with other participant reflections, this relationship can also be explored in a contrasting direction, as it has been suggested by Jones et al. (2014) that "differences in temperament and emotion result from experiences with stuttering" (p.122).

Parent self-regulation

Parent self-regulation has been identified as crucial in the success of parenting interventions, and emerged as a prominent theme in the parent interviews (Sanders & Mazzucchelli, 2013). Bandura (1977) indicates that specific self-efficacy beliefs, a component of self-regulation, guides a person's behavior and predicts how well a task is performed. Given the key role that parents play as active agents of change in direct or indirect stuttering interventions (Druker, Hennessey, Hennessey et al., 2019, 2019c; Franken & Putker-de Bruijn, 2007; Millard et al., 2008; Onslow & O'Brian, 2013), the importance of parent self-regulation in the context of stuttering management needs to be considered. The positive shift in parent-self regulation reported by parents in the current study may be important for their own improved emotional regulation, their interactions with their children, as well as their capacity and confidence to problem solve the management of their children's behavior and stuttering issues concurrently (Barros, Goes, & Pereira, 2015).

Family dynamics

Parents reported that the integrated stuttering treatment and behavioral self-regulation program supported different considerations of their family dynamics. Improved parent-child relationships, for example, was reflected on strongly in the interviews, and has implications for stuttering treatment. Improvements in parent-child relationships and interactions are likely to increase the child's motivation to follow instructions (Bor et al., 2002), and may consequently result in the child potentially being more responsive to their parents' suggestions and strategies to facilitate fluency. Furthermore, the quality of the parent-child relationship has consequences for child mental well-being, with positive and nurturing parent-child relationships found to promote good psychological functioning "across life up to and including the seventh decade" (Stafford, Kuh, Gale, Mishra, & Richards, 2016, p. 335).

In the context of stuttering, the DCM encourages parents to identify factors in the child's environment that may be affecting the child's speech. Although the home environment has not been found to significantly predict stuttering severity (Kraft et al., 2014, 2018), one parent reported that her child's stutter was exacerbated when he kept trying to interrupt and stop his parents from arguing. Furthermore, in light of the principles of the stuttering treatment implemented in this study, whereby support for fluency is provided throughout daily conversation interactions, parents working more consistently as a team considered themselves to be more effective in collectively implementing strategies to best support their child's fluency throughout the day.

Among a broad range of consequences, siblings of children with ADHD have experienced differential parent treatment (King et al., 2016), and have been identified with increased relationship problems and higher levels of conflict than typical child-sibling dyads (Mikami & Pfiffner, 2008). Such effects may have also been experienced by siblings in the current study, given that all children who stutter met criteria for concomitant elevated ADHD symptoms. Siblings of children who stutter have exhibited strong emotions about having a sibling who stutters, and have also expressed a desire to be more involved in the therapy process (Beilby, 2014). In this light, the beneficial flow-on effect to siblings reflected on in the study's interviews, may be important to consider in future holistic intervention approaches.

Positive parent perceptions of the integrated program

The valuable parent reflections discussed the perceived importance of early intervention, and captured the apparent alleviation of stress, despair and helplessness when provided with support for their children's developmental and behavioral difficulties close to onset (Millard & Davis, 2016; Plexico & Burruss, 2012). Therapist support playing a crucial role in participant engagement and success was spoken about by a number of participants, supporting literature which emphasizes the importance of therapeutic alliance in healthcare program engagement and treatment outcomes (Manning & DiLollo, 2018; Easterbrook & Meehan, 2017). Nearly half of the participants reflected that ease of the delivery of the program facilitated their involvement and completion of the program. For example, SM reported that telehealth delivery of the program was a key enabler in her program completion, as she had previously

been recommended to attend a Triple P program for her daughter's behavioral problems, and had declined due to inaccessibility. This aligns with Mazzucchelli and Sanders (2010) who described the importance of flexible delivery to meet the diverse needs of consumers, and concluded that rigid program adherence "may not actually be necessary or lead to optimal delivery" (p. 239).

Parents of children who stutter have previously found it challenging to accommodate daily stuttering treatment practice sessions into the "busy family schedule" (Van Eerdenbrugh, Packman, O'Brian, & Onslow, 2018, p. 1267). By contrast, the current study's naturalistic approach to stuttering treatment (Druker, Hennessey, Hennessey et al., 2019a, 2019c) was perceived as a positive feature of the intervention program, supporting treatment engagement and implementation of techniques beyond the clinic environment. Perceived impracticality of an intervention can act as a barrier to its uptake and effectiveness (Mazzucchelli et al., 2010). Therefore, parents perceiving the integrated program techniques as practical, naturalistic, and easy to administer, supports the functional and social validity of the CUSP intervention for the future.

Of interest, parents did not reflect on either treatment component of the integrated program in isolation and instead focused on how the two components integrated seamlessly together. Parents reflected that when their child was stuttering less, they were happier communicators and less feelings of anger and frustration occurred. This worked in the reverse as well. For example, one parent (AM) reflected that implementing house rules as part of the Triple P component "takes the pressure off day to day negative interactions, so interactions are a lot more positive, there's less complaining and whining, and less snitching. This means that [our child] is naturally stuttering less, because the stuttering happened most when she was whining or trying to snitch on her brother".

The acceptability of intervention principles is imperative, as perceived lack of acceptability can act as a barrier to parent engagement and uptake of an intervention (Mazzucchelli et al., 2010). Furthermore, parents who perceive strategies or principles as "unacceptable or impractical may not attempt potentially useful and effective strategies" (Mazzucchelli et al., 2010, p. 118). In the current study, the integrated program was perceived as beneficial by every participant.

Limitations

The data from this study do not establish the effectiveness of the integrated stuttering treatment and behavioral self-regulation program, since no empirical outcome measures are reported (quantitative treatment outcomes is the focus of an associated paper, Druker, Hennessey et al., 2019). Furthermore, in some instances, it is difficult to tease out parent comments that might have occurred if the child's fluency had improved in isolation, compared to changes resultant from the possible treatment interaction. In the associated intervention study (Druker, Hennessey et al., 2019), 19 parents were randomly assigned to the group in which the integrated program was administered. From this larger treatment group, eight parents who met eligibility criteria for the current study were recruited. A larger sample may have provided more varied reflections of the integrated program, although analyses revealed that saturation was reached with the current cohort. The sample is relatively homogenous with all parents being Caucasian and of a similar socioeconomic demographic, thus potentially limiting the generalizability of findings to the broader population. All participants interviewed were mothers. Future research exploring experiences of other partners is recommended.

Conclusion

This study explored the experiences of eight different parents with an integrated stuttering treatment and behavioral self-regulation program designed to support their child who stuttered who also had concomitant self-regulation challenges. The qualitative research approach allowed the authors to capture the experiences and reflections of these parents, beyond what would have been achieved with quantitative measures. Significant themes of emotional impact on parents, child self-regulation, links between stuttering and behavior, parent self-regulation, family dynamics, as well as positive perceptions of the integrated program emerged strongly and consistently as major qualitative themes. Conclusions provide support for the social validity of an integrated stuttering and self-regulation intervention, as well as the perceived benefits of the program for future families with children who stutter and have elevated behavioral challenges.

Conflict of Interest

The authors report no conflicts of interest.

Appendix A

Question used to guide qualitative interviews

1 How did you feel when your child started stuttering?
2 How do/did you manage your child's stuttering?
3 What, if anything, exacerbates/ed your child stutter?
4 Has your child's stuttering ever affected you emotionally?
5 Have your child's behavior problems/self-regulation skills ever affected you emotionally?
6 Has your child's stuttering ever affected your relationship with your child? If so, how?
7 Has your child's behavior problems/ self-regulation skills ever affected your relationship with your child? If so, how?
8 How confident did you feel in managing your child's behavior problems?
9 Before starting the program, did you notice any relationship to exist, between your child's stuttering and his/her behavior or self-regulation skills?
10 Do you feel the program has any benefits or disadvantages to parents of children who stutter? Please describe.
11 Are there any further thoughts, comments or reflections that you'd like to share with us?

References

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioral change. *Psychological Review, 84*, 181–215.
Barbour, R. S. (2001). Checklists for improving rigour in qualitative research: a case of the tail wagging the dog? *British Medical Journal, 322*, 1115–1117.
Barkley, R. (2013). *Taking charge of ADHD, third edition : The complete, authoritative guide for parents* (3 ed.). New York, NY: Guildford Publications.
Barros, L., Goes, A. R., & Pereira, A. I. (2015). Parental self-regulation, emotional regulation and temperament: Implications for intervention. *Estudos De Psicologia, 32*(2), 295–306. https://doi.org/10.1590/0103-166X2015000200013.
Beilby, J. (2014). Psychosocial impact of living with a stuttering disorder: Knowing is not enough. *Seminars in Speech and Language, 35*(2), 132–143. https://doi.org/10.1055/s-0034-1371756.
Bertakis, K. D., & Azari, R. (2011). Patient-centred care is associated with decreased health care utilization. *Journal of the American Board of Family Medicine, 24*(3), 229–239.
Binns, A., Hutchison, L., & Oram Cardy, J. (2019). The speech-language pathologist's role in supporting the development of self-regulation: A review and tutorial. *Journal of Communication Disorders, 78*, 1–17. https://doi.org/10.1016/.jcomdis.2019.12.005.
Bor, W., Sanders, M., & Markie-Dadds, C. (2002). The effects of the Triple P-Positive Parenting Program on preschool children with co-occurring disruptive behavior and attentional/hyperactive difficulties. *Journal of Abnormal Child Psychology, 30*(6), 571–587. https://doi.org/10.1023/A:1020807613155.
Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77–101. https://doi.org/10.1191/1478088706qp063oa.
Brown, M., McIntyre, L., Crnic, K., Baker, B., & Blacher, J. (2011). Preschool children with and without developmental delay: risk, parenting, and child demandingness. *Journal of Mental Health Research in Intellectual Disabilities, 4*(3), 206–226. https://doi.org/10.1080/19315864.2011.596990.
Campbell, S. B. (2000). *Attention-deficit/hyperactivity disorder: a developmental viewHandbook of developmental psychopathology* (2 ed.). New York: Kluwer Academic/Plenum383–401.
Connery, A., McCurtin, A., & Robinson, K. (2019). The lived experience of stuttering: a synthesis of qualitative studies with implications for rehabilitation. *Disability and Rehabilitation, 29*, 1–11. https://doi.org/10.1080/09638288.2018.1555623.
Conture, E., Kelly, E., & Walden, T. (2013). Temperament, Speech and Language: An Overview. *Journal of Communication Disorders, 46*(2), 125–142. https://doi.org/10.1016/j.jcomdis.2012.11.002.
Danforth, J. S., Barkley, R. A., & Stokes, T. F. (1991). Observations of parent-child interactions with hyperactive children: Research and clinical implications. *Journal of the American Academy of Child and Adolescent Psychiatry, 40*, 508–515. https://doi.org/10.1016/0272-7358(91)90127-G.
De Sonneville-Koedoot, C., Stolk, E., Rietveld, T., & Franken, M.-C. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PLoS One, 10*(7), 1–17. https://doi.org/10.1371/journal.pone.0133758.
Deater-Deckard, K. (2014). Family matters: Intergeneration and interpersonal processes of executive function and attentive behaviour. *Current Directions in Psychological Science, 23*, 230. https://doi.org/10.1177/0963721414531597.
Delaney, L. (2018). Patient-centred care as an approach to improving health care in Australia. *The Australian Journal of Nursing Practice, Scholarship and Research, 25*(1), 119–123. https://doi.org/10.1016/j.colegn.2017.02.005.
Donaher, J., & Richels, C. (2012). Traits of Attention Deficit/Hyperactivity Disorder in school-age children who stutter. *Journal of Fluency Disorders, 37*(4), 242–252. https://doi.org/10.1016/j.jfludis.2012.08.002.
Druker, K., Hennessey, N., Mazzucchelli, T., & Beilby, J. (2019a). *An evaluation of an integrated stuttering and parent-administered self-regulation program for early developmental stuttering disorders.* Manuscript in preparation.
Druker, K., Hennessey, N., Mazzucchelli, T. G., & Beilby, J. (2019b). Elevated attention deficit hyperactivity disorder symptoms in children who stutter. *Journal of Fluency Disorders, 59*, 80–90. https://doi.org/10.1016/j.jfludis.2018.11.002.
Druker, K., Mazzucchelli, T. G., & Beilby, J. (2019). An evaluation of integrated fluency and resilience program for early developmental stuttering disorders. *Journal of Communication Disorders, 78*, 69–83. https://doi.org/10.1016/j.jcomdis.2019.02.002.
DuPaul, G. J., McGoey, K. E., Eckert, T. L., & VanBrakel, J. (2001). Preschool children with attention-deficit/hyperactivity disorder: Impairments in behavioural, social and school functioning. *Journal of American Academy of Child and Adolescent Psychiatry, 40*, 508–515. https://doi.org/10.1097/00004583-200105000-00009.
Easterbrook, C. J., & Meehan, T. (2017). The therapeutic relationship and cognitive behavioural therapy: A case study of an adolescent girl with depression. *The European Journal of Counselling Psychology, 6*(1), 1–24. https://doi.org/10.5964/ejcop.v6i1.85.
Eggers, K., De Nil, L., & Van Den Bergh, B. (2013). Inhibitory control in childhood stuttering. *Journal of Fluency Disorders, 38*, 1–13. https://doi.org/10.1016/j.jfludis.2012.10.001.
Eggers, K., De Nil, L., & Van den Bergh, B. (2013). Temperament dimensions in stuttering and typically developing children. *Journal of Fluency Disorders, 35*, 355–372. https://doi.org/10.1016/j.jfludis.2010.10.004.
Eggers, K., De Nil, L. F., & Van Den Bergh, B. R. (2012). The efficiency of attentional networks in children who stutter. *Journal of Speech, Language and Hearing Research, 55*(3), 946–959. https://doi.org/10.1044/1092-4388(2011/10-0208).
Eisenberg, N., & Sulik, M. J. (2012). Emotion-related self-regulation in children. *Teaching of Psychology, 39*(1), 77–83. https://doi.org/10.1177/0098628311430172.
Ekman, I., Wolf, A., Olsson, L., Taft, C., Dudas, K., Schaufelberger, M., ... Swedberg, K. (2012). Effects of person-centred care in patients with chronic heart failure: The PCC-HF study. *European Heart Journal, 33*(9), 1112–1119.
Ellis, B., & Nigg, J. (2009). Parenting practices and attention-deficit/hyperactivity disorder: new findings suggest partial specificity of effects. *Journal of American Academy of Child and Adolescent Psyhchiatry, 48*(2), 146–154. https://doi.org/10.1097/CHI.0b013e31819176d0.
Eyberg, S. (1988). Parent-child interaction therapy: Integration of traditional and behavioral concerns. *Child and Behavior Therapy, 10*, 33–46. https://doi.org/10.1300/J019v10n01_04.
Felsenfeld, S., Van Beljsterveldt, C., & Boomsma, D. I. (2010). Attentional regulation in young twins with probable stuttering, high nonfluency, and typical fluency. *Journal of Speech, Language, and Hearing Research, 53*(5), 1147–1166. https://doi.org/10.1044/1092-4388(2010/09-0164).

Franken, M. C., & Putker-de Bruijn, D. (2007). *Restart-DCM Method. Treatment protocol developed within the scope of the ZonMW project Cost-effectiveness of the Demands and Capacities Model based treatment compared to the Lidcombe programme of early stuttering intervention: Randomised trial.*

Gross, H., Shaw, D., & Moilanen, K. (2008). Reciprocal associations between boys' externalizing problems and mothers' depressive symptoms. *Journal of Abnormal Child Psychology, 36*, 693–709. https://doi.org/10.1007/s10802-008-9224.

Guest, G., Bunce, A., & Johnson, L. (2006). How many interviews are enough? An experiment with data saturation and variability. *Field Methods, 18*(1), 59–82. https://doi.org/10.1177/1525822X05279903.

Haskett, M. E., Armstrong, J., Neal, S., & Aldianto, K. (2018). Perceptions of Triple P-Positive Parenting Program seminars among parents experiencing homelessness. *Journal of Child and Family Studies, 27*, 1957–1967. https://doi.org/10.1007/s10826-018-1016-5.

Hoath, F., & Sanders, M. (2002). A feasibility study of Enhanced Group Triple P - positive parenting program for parents of children with Attention-deficit/hyperactivity Disorder. *Behaviour Change, 19*(4), 191–206.

Houlding, C., Schmidt, F., Stern, S. B., Jamieson, J., & Borg, D. (2012). The perceived impact and acceptability of Group Triple P Positive Parenting Program for Aboriginal parents in Canada. *Children and Youth Services Review, 34*(12), 2287–2294. https://doi.org/10.1016/j.childyouth.2012.08.001.

Jones, R., Choi, D., Conture, E., & Walden, T. (2014). Temperament, emotion and childhood stuttering. *Seminars in Speech and Language, 35*(2), 114–131. https://doi.org/10.1055/s-0034-1371755.

Karrass, J., Walden, T., Conture, E., Graham, C., Arnold, H., Hartfield, K., ... Schwenk, K. (2006). Relation of emotional reactivity and regulation to childhood stuttering. *Journal of Communication Disorders, 39*, 402–423. https://doi.org/10.1016/j.jcomdis.2005.12.004.

King, K., Alexander, D., & Seabi, J. (2016). Siblings' perceptions of their ADHD-diagnosed sibling's impact on the family system. *International Journal of Environmental Research and Public Health, 13*, 910–920. https://doi.org/10.3390/ijerph13090910.

Kraft, S. J., Ambrose, N., & Chon, H. (2014). Temperament and environmental contributions to stuttering severity in children: the role of effortful control. *Seminars in Speech and Language, 35*(2), 80–94. https://doi.org/10.1055/s-0034-1371753.

Kraft, S. J., Beilby, J., & Lowther, E. (2018). The role of effortful control in stuttering severity in children: Australian replication study. *American Journal of Speech-Language Pathology,* 1–15. https://doi.org/10.1044/2018_AJSLP-17-0097.

Langevin, M., Packman, A., & Onslow, M. (2010). Parent perceptions of the impact of stuttering on their preschoolers and themselves. *Journal of Communication Disorders, 43*(5), 407–423. https://doi.org/10.1016/j.jcomdis.2010.05.003.

Manning, W. H., & DiLollo, A. (2018). *Clinical decision making in fluency disorders.* San Diego, CA: Plural Publishing Incorporated.

Mazzucchelli, T. G., & Sanders, M. R. (2010). Facilitating practitioner flexibility within an empirically supported intervention: Lessons from a system of parenting support. *Clinical Psychology Science and Practice, 17*(3), 238–252. https://doi.org/10.1111/j.1468-2850.2010.01215.

Mazzucchelli, T. G., Studman, L. J., Whittingham, K., & Sofronoff, K. (2010). Comments on a study of social validity of the developmental disability parent training program Stepping Stones Triple P. *Life Span and Disability, XIII*(2), 109–128. doi:Retrieved from https://espace.library.uq.edu.au/view/UQ:237181.

Mikami, A. Y., & Pfiffner, L. J. (2008). Sibling relationships among children with ADHD. *Journal of Attention Disorders, 11*, 482–492. https://doi.org/10.1177/1087054706295670.

Millard, S., Nicholas, A., & Cook, F. (2008). Is parent-child interaction therapy effective in reducing stuttering? *Journal of Speech, Language and Hearing Research, 51*(3), 636–650.

Millard, S. K., & Davis, S. (2016). The Palin Parent Rating Scales: Parents' perspectives of childhood stuttering and its impact. *Journal of Speech, Language and Hearing Research, 59*, 950–963. https://doi.org/10.1044/2016_JSLHR-S-14-0137.

Minnes, P., Perry, A., & Weiss, J. (2015). Predictors of distress and well-being in parents of young children with developmental disabilities: the importance of parent perceptions. *Journal of Intellectual Disability Research, 59*(6), 551–560. https://doi.org/10.1111/jir.12160.

Moffitt, T., Arseneault, L., Belsky, D., Dickson, N., Hancox, R. J., Harrington, H., ... Caspi, A. (2011). Gradient of childhood self-control predicts health, wealth, and public safety. *Proceedings of the National Academy of Sciences of the United States of America, 108*(7), 2693–2698. https://doi.org/10.1073/pnas.1010076108.

Neece, C., Green, S., & Baker, B. (2012). Parenting stress and child behavior problems: A transactional relationship across time. *American Journal on Intellectual and Developmental Disabilities, 117*, 48–66. https://doi.org/10.1352/1944-7558-117.1.48.

Onslow, M., & O'Brian, S. (2013). Management of childhood stuttering. *Journal of Paediatrics and Child Health, 49*(2), 112–115. https://doi.org/10.1111/jpc.12034.

Patterson, G. R. (2005). The next generation of PMTO models. *Behavioral Therapist, 28*(2), 27–33.

Peters, K., & Jackson, D. (2008). Mothers' experiences of parenting a child with attention deficit hyperactivity disorder. *Journal of Advanced Nursing, 65*, 62–71. https://doi.org/10.1111/j.1365-2648.2008.04853.x.

Plexico, L., & Burrus, E. (2012). Coping with a child who stutters: a phenomenological analysis. *Journal of Fluency Disorders, 37*, 275–288. https://doi.org/10.1016/j.jfludis.2012.06.002.

Riley, G., & Riley, J. (2000). A revised component model for diagnosing and treating children who stutter. *Contmeporary Issues in Communication Sciences and Disorders, 27*, 188–199.

Sanders, M. (2003). Triple P – Positive Parenting Program: A population approach to promoting competent parenting. *Australian e-Journal for the Advancement of Mental Health, 2*(3), 127–143. https://doi.org/10.5172/jamh.2.3.127.

Sanders, M., Kirby, J., Tellegen, C., & Day, J. (2014). The Triple P-Positive Parenting Program: A systematic review and meta-analysis of a multi-level system of parenting support. *Clinical Psychology Review, 34*(4), 337–357. https://doi.org/10.1016/j.cpr.2014.04.003.

Sanders, M. R., & Mazzucchelli, T. G. (2013). The promotion of self-regulation through parenting interventions. *Clinical Child and Family Psychology Review, 16*(1), 1–17. https://doi.org/10.1007/s10567-013-0129-z.

Sanders, M. R., & Mazzucchelli, T. G. (2018). *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system.* New York, NY: Oxford University Press.

Shafiei, B., Faramarzi, S., Abedi, A., Dehqan, A., & Scherer, R. (2019). Effects of the Lidcombe Program and Parent-Child Interaction Therapy on stuttering reduction in preschool children. *Folia Phoniatrica et Logopaedica, 71*, 29–41. https://doi.org/10.1159/000493915.

Smith, A., & Weber, C. (2016). Childhood stuttering: Where are we and where are we going? *Seminars in Speech and Language, 37*(4), 291–297. https://doi.org/10.1055/s-0036-1587703.

Smith, A., & Weber, C. (2017). How stuttering develops: the multifactorial dynamic systems pathways theory. *Journal of Speech, Language, and Hearing Research, 60*, 2483–2505. https://doi.org/10.1044/2017_JSLHR-S-16-0343.

Sonney, J., & Insel, K. (2016). Reformulating the common sense model of self-regulation: toward parent-child shared regulation. *Nursing Science Quarterly, 29*(2), 154–159. https://doi.org/10.1177/0894318416630091.

Sonuga-Barke, E. J. J., Daley, D., Thompson, M., Laver-Bradbury, C., & Weeks, A. (2001). Parent-based therapies for preschool attention-deficit/hyperactivity disorder: A randomised, controlled trial with a community sample. *Journal of American Academt of Child and Adolescent Psyhciatry, 40*, 402–408. https://doi.org/10.1097/00004583-200104000-00008.

Stafford, M., Kuh, D. L., Gale, C. R., Mishra, G., & Richards, M. (2016). Parent-child relationships and offspring's positive mental wellbeing from adolescence to early older age. *The Journal of Positive Psychology, 11*(3), 326–337. https://doi.org/10.1080/17439760.2015.1081971.

Starkweather, W. C. (1987). *Fluency and stuttering.* Englewood Cliffs, N.J: Prentice-Hall.

Starkweather, W. C., Ridener Gottwald, S., & Halfond, M. (1997). *Stuttering prevention: a manual for parents.* 11.3.1997 Retrieved from https://www.mnsu.edu/comdis/kuster/Parents/starkweather.html.

Tsukayama, E., Toomey, S. L., Faith, M. S., & Duckworth, A. L. (2010). Self-control as a protective factor against overweight status in the transition from childhood to adolescense. *Archives of Pediatrics and Adolescent Medicine, 164*, 631–635. https://doi.org/10.1001/archpediatrics.2010.97.

Ullsperger, J., Nigg, T., & Nikolas, M. (2016). Does child temperament play a role in the association between parenting practices and child attention deficit/hyperactivity disorder? *Journal of Abnormal Child Psychology. 44*, 167–178. https://doi.org/10.1007/s10802-015-9982-1.

Van Eerdenbrugh, S., Packman, A., O'Brian, S., & Onslow, M. (2018). Challenges and strategies for speech-language pathologists using the Lidcombe program for early stuttering. *American Journal of Speech-Language Pathology, 27*, 1259–1272. https://doi.org/10.1044/2014_JSLHR-S-13-0090.

van Steijn, D., Oerlemans, A., van Aken, M., Buitelaar, J., & Rommelse, N. (2014). The reciprocal relationship of ASD, ADHD, depressive symptoms and stress in parents of children with ASD and/or ADHD. *Journal of Autism and Developmental Disorders, 44*, 1064–1076. https://doi.org/10.1007/s10803-013-1958-9.

Vanryckeghem, M., Brutten, G. J., & Hernandez, L. M. (2005). A comparative investigation of the speech-associated attitude of preschool and kindergarten children who do and do not stutter. *Journal of Fluency Disorders, 30*(4), 307–318. https://doi.org/10.1016/j.jfludis.2005.09.003.

Walden, T., Frankel, C., Buhr, A., Johnson, K., Conture, E., & Karrass, J. (2012). Dual diathesis-stressor model of emotional and linguistic contributions to developmental stuttering. *Journal of Abnormal Child Psychology, 40*(4), 633–644. https://doi.org/10.1007/s10802-011-9581-8.

Webster-Stratton, C. (1998). Preventing conduct problems in Head Start children: Strengthening parenting competencies. *Journal of Consulting and Clinical Psychology, 66*(5), 715–730. https://doi.org/10.1037/0022-006X.66.5.715.

Yaruss, J. S., Coleman, C., & Hammer, D. (2006). Treating preschool children who stutter: Description and preliminary evaluation of a family-focused treatment approach. *Language, speech, and hearing services in schools, 37,* 118–136 doi:0161-1461/06/3702-0118.

Yaruss, J. S., & Quesal, R. W. (2004). Stuttering and the International Classification of Functioning, Disability, and Health (ICF): An update. *Journal of Communication Disorders, 37*(1), 35–52.

Kerianne C. Druker is a PhD candidate at Curtin University, Perth, Western Australia. She is a certified practising speech pathologist. Her clinical and research areas of interest include treatment of fluency disorders in paediatric populations and examining the role that self-regulation plays in early-onset stuttering.

Trevor G. Mazzucchelli is a registered clinical psychologist who has specialised in parenting interventions for families of children with behaviour disorders. He is also a senior lecturer in clinical psychology at Curtin University, Perth, Western Australia.

Neville W. Hennessey is a lecturer in the School of Occupational Therapy, Social Work and Speech Pathology at Curtin University, Perth, Western Australia. His current research interests include stuttering, speech disorders in children and adults, language and literacy development and disorders, and psycholinguistics.

Janet M. Beilby is an Associate Professor of Stuttering Disorders in the School of Occupational Therapy, Social Work and Speech Pathology at Curtin University, Perth, Western Australia. Her current research interests include parent-administered early treatment initiatives, adult stuttering treatment programs and the psychosocial impact of stuttering disorders.

Chapter 6

Discussion

Chapter 6: Discussion

The overarching aim of the current body of work was to explore the impact of self-regulation issues, manifesting in elevated ADHD symptoms, on stuttering treatment outcomes. In order to do this, the impact of these self-regulation issues on stuttering treatment outcomes was investigated retrospectively and prospectively. Consideration was given to subsequently address these symptoms through a novel, integrated stuttering and self-regulation intervention. The novel program was evaluated using mixed quantitative and qualitative research to capture the potential benefits and acceptability of the approach for this population. The PhD research project comprised a series of three separate, but interrelated, studies.

Study 1 employed a retrospective clinical audit of 185 CWS who presented to clinic for stuttering treatment. Key research aims in Study 1 included identifying the proportion of young children with a clinical diagnosis of a developmental stuttering disorder who also had elevated ADHD symptoms. The impact of these symptoms on stuttering treatment responsiveness was determined. This study identified a significant proportion of CWS to present with self-regulation issues, and evaluated the impact of these symptoms on stuttering treatment responsiveness. Considering patient comorbidities has long been a crucial component of holistic care in the medical field (Eisele et al., 2018). The large proportion of CWS who were found to present with comorbid elevated ADHD symptomology, as well as the demonstrated negative impact of these symptoms on stuttering treatment responsiveness, provided a sound rationale for their consideration in holistic stuttering management.

Building on the findings from the initial investigation, Study 2 implemented a three-pronged controlled clinical trial to evaluate the effectiveness of the novel, integrated stuttering and self-regulation intervention on stuttering treatment outcomes. This intervention was found to be successful in improving stuttering treatment outcomes for CWS with self-regulation issues. This is promising in the context of the major negative psychosocial consequences of living with a chronic stuttering disorder (e.g., Carter et al., 2017).

In conjunction with this clinical trial whereby quantitative research methodology was used to evaluate treatment effectiveness, the final study (Study 3) of this PhD research employed qualitative research methodology to gain parent reflections and experiences with the novel program. These reflections augmented

findings captured using quantitative methodology only (Zhang, 2014). Of particular interest for future clinical consideration were the parent reported emotional consequences of having CWS with co-occurring ADHD symptoms, such as feelings of frustration, stress, and concern for their children's future. In addition, the reported links between stuttering and behavioural self-regulation in daily life activities further validated the possible psychosocial benefits of this program for future clinical practice.

Key highlights and limitations which evolved from the major research questions are explored below.

Elevated ADHD symptoms: Proportion in CWS and Impact on Stuttering Treatment Outcomes

Identifying the proportion of CWS with elevated ADHD symptoms, as well as the possible impact of these symptoms on stuttering treatment outcomes, was considered important to ascertain if these symptoms should be considered and addressed in stuttering management. Previous research has examined the comorbidity of diagnosed ADHD in the stuttering population, and has found the proportion of ADHD in people who stutter to be only slightly higher than the people who do not stutter (Blood et al., 2003). However, research comparing the presence of elevated ADHD symptoms or ADHD-like traits in people who stutter and those who do not, has consistently found attention difficulties to be elevated in those who stutter (Alm & Risberg, 2007; Donaher & Richels 2012). One previous study investigated the proportion of CWS with elevated ADHD symptoms, as opposed to clinically diagnosed ADHD (Donaher & Richels, 2012). The authors found 57% of CWS to present with these elevated symptoms, out of a sample size of 36 children.

The results of Study 1 determined that 49.7% of 185 CWS presented with subclinical behavioural self-regulation challenges, measured by elevated ADHD symptoms on the ADHD Rating Scale (McGoey et al., 2007; Power et al., 2001). A limitation of this study was the retrospective nature of the investigation, which resulted in a three-month delay between discharge from stuttering treatment and parent completion of the ADHD rating scale. Although a notable proportion of behavioural problems persist between 2 to 4.5 years of age (D'Souza et al., 2019), it is acknowledged that behaviour problems are also known to fluctuate in these preschool years (Basten et al., 2016; D'Souza et al., 2019). This limitation was addressed by the prospective nature of the clinical intervention trial implemented in

Study 2. Findings were successfully replicated in this prospective trial, as there was a similar proportion of CWS who met criteria for elevated ADHD symptoms at pre-treatment (i.e., 36 out of 76 children, 47%). It is worth noting that 16 out of the 17 children who met criteria for elevated ADHD symptoms at pre-treatment, who received standard stuttering treatment only, continued to meet these criteria at post-treatment, providing evidence of the relatively stable nature of these symptoms if not addressed. Of the 19 children who met criteria for elevated ADHD symptoms at pre-treatment, and who received the integrated intervention, nine no longer satisfied these criteria at post-treatment (while the remainder still demonstrated a reduction in ADHD symptoms).

The large proportion of CWS, who were found to have co-occurring elevated ADHD symptoms, is consistent with previous research (Donaher & Richels, 2012), and was replicated by two independent larger samples in the current body of research. Study 1 also revealed that the presence of attentive and hyperactive/impulsive symptoms predicted poorer responsiveness to stuttering treatment. Specifically, CWS who met criteria for elevated ADHD symptomology required, on average, a 24% increase in clinical intervention sessions compared to CWS who did not present with this comorbid symptomology. These findings were replicated in Study 2, when the number of stuttering intervention sessions required in the two treatment groups who only received stuttering treatment was compared. CWS with elevated symptoms who only received stuttering treatment required an additional 32% of stuttering treatment intervention time, compared to CWS without these symptoms. These findings align with an earlier study by Riley and Riley (2000), whereby the presence of attending difficulties was associated with a lack of stability in stuttering reduction at 2- to 4-year follow-up.

In terms of post-treatment stuttering severity, Study 1 showed no statistically significant differences between CWS with and without elevated ADHD symptoms (eADHD). However, the stuttered speech severity in the group of CWS without eADHD satisfied the criteria for normal fluency at post-treatment (stuttered speech severity of 2%SS or less), while this was not achieved in the group of children with eADHD. This indicates the mean stuttered speech severity for the group of CWS with eADHD was above the cut-off for normal fluency following treatment, and provides some evidence that ADHD symptoms did in fact influence the outcomes of stuttering treatment.

In Study 2, however, CWS with eADHD who received the CUSP were observed to have a significantly higher %SS at post-treatment and follow-up, compared to CWS without the comorbid symptoms who received the equivalent stuttering intervention program. Although statistically significant differences in post-treatment stuttered speech severity were not present between groups of CWS with and without comorbid eADHD symptoms in Study 1, such differences may have been present if a follow-up or maintenance period was included. This hypothesis is based on the successful impact of the integrated self-regulation and stuttering treatment program on fluency outcomes demonstrated in Study 2, with larger effects demonstrated at follow-up compared to post-treatment; suggesting the impacts of self-regulation skills on stuttering manifestation may not be immediately evident following a treatment block.

Effectiveness of Intervention

The Integrated Stuttering and Self-Regulation Program

The novel integrated stuttering and self-regulation program was implemented with the primary aim of improving stuttering treatment outcomes for CWS challenged with eADHD. This program was judged to be effective for this group of CWS, when considering their stuttering treating outcomes. A key finding was that 95% of these CWS with eADHD who received the integrated program met criteria for normal fluency at follow-up. This was comparable to the percentage of CWS without comorbid ADHD symptoms who met this criteria. This 95% responsiveness rate is higher than the average 75% of CWS who have had their stuttering disorders remediated following other published early stuttering interventions (e.g., De Sonneville-Koedoot et al., 2015), and is a particularly significant proportion for a group of children who tend not to show the same level of remediation following intervention, due to comorbid symptoms (Riley & Riley, 2000). It would be worthwhile determining if this high responsiveness rate can be replicated in future research.

Outcomes from the novel intervention program provide support for tailoring interventions to address comorbid issues or unique characteristics within the profiles of CWS, to maximise their chances of ultimately recovering from stuttering. Self-regulation has shown to be a particularly potent issue to address in developmental stuttering, with no other known comorbid factor showing such an influence on stuttering treatment outcomes to date. The individualised treatment strategies align

with the recommended future directions in a recent review of published paediatric stuttering interventions, whereby the importance of individualised intervention approaches was emphasised (Donaghy & Smith, 2016).

Our findings also suggested that the impact of improved self-regulation skills on the fluency trajectories of these children may continue beyond the three-month follow-up data collection time in the study. This assumption is drawn from the finding that the benefits of self-regulatory improvements appeared to have greater effects on stuttered speech severity at follow-up.

Beyond the improvements to the young child's fluency, this integrated stuttering and self-regulation intervention also yielded large and significant improvements in the child's behaviour outcomes as well as the parenting practices and parental/family adjustment within the family constellation. The functional impacts of having a child with diagnosed ADHD are well documented, in terms of the possible impacts on parental mental health and familial harmony and relationships (Barkley, 2013; Lifford et al., 2008; Mikami & Pfiffner, 2008; Van Steijn et al., 2014). Therefore, the presence of eADHD in a large subgroup of CWS are likely to mirror some of these functional impacts, albeit to a lesser degree, given the subclinical nature of these symptoms. The possible negative impacts of having a child who stutters with co-occurring behavioural regulation issues were demonstrated through parent reflections gathered in Study 3, as well as the pre-treatment differences in psychosocial measures between the elevated and non-elevated groups, including on the SDQ. The most prominent theme in the parent interviews was the emotional impact they experienced as a result of having a child with these comorbid issues. Therefore, positive improvements in parenting practices and family adjustment was beneficial for the children and their families from a psychosocial perspective, beyond the benefits to improving stuttering treatment outcomes (Langevin et al., 2010; Moffitt et al., 2011; Plexico & Burrus, 2012).

Curtin University Stuttering Program

The CUSP was the stuttering intervention implemented in Studies 1 and 2 of this PhD project. As such, the qualitative comments relating to fluency management in Study 3, reflected, in part, parents' experiences with the CUSP. The stuttering treatment used in this research yielded significant reductions in the stuttered speech severity for these young children who presented for clinical intervention and support. The research demonstrated positive treatment outcomes and large effect sizes,

comparable to those documented in published treatments of early stuttering intervention, including the Lidcombe Program and RESTART-DCM (De Sonneville-Koedoot et al., 2015). Both retrospective and prospective clinical data were used to evaluate the outcomes of the CUSP. Retrospective data (Study 1) were collected from 185 children, and prospective pre, post, and follow-up data (Study 2) were gathered from 57 children. The children who received the CUSP integrated with EBPS were excluded from the latter total sample size due to possible effects of the EBPS component on the stuttering treatment responsiveness. The large sample sizes and the significant and successful fluency outcomes underscore the fluency gains that resulted from the CUSP intervention.

There are preliminary indications from this body of research that the CUSP may be more cost effective than previously published early stuttering treatment programs, given similar stuttering treatment outcomes resulted with less clinical intervention time. The mean number of sessions required in the CUSP was 12, compared to 22 in the Lidcombe Program and 20 in the DCM. In addition, the mean number of treatment hours required in the Lidcombe Program was 19.6, 18.0 in the DCM and 7.3 in the CUSP.

The treatment efficacy, clinic time and cost effectiveness of the CUSP provides endorsement for future research to compare the effectiveness of the CUSP to other published preschool stuttering interventions (e.g., the Lidcombe Program or RESTART-DCM) in a randomised prospective controlled clinical trial. Furthermore, long-term follow-up of children who receive the CUSP, compared to other treatment programs, would provide information about the durability of fluency improvements across treatment programs. Such proposed future research is important to determine the most effective and efficient approach to addressing stuttering in younger children.

An additional outcome that supports the effectiveness of the integrated program, and the CUSP, is the low attrition rate in this research. Attrition rates have widely been used as a measure of the acceptability of clinical health interventions (Sekhon et al., 2017). Previous research has reported low recovery rates from stuttering post 12 months onset (Carey et al., 2020a; Reilly et al., 2013) and attrition rates as high as 43% in previous preschool stuttering intervention programs (e.g., Trajkovski et al., 2019). There have been reports of significant numbers of children who enter a clinical intervention trial who do not complete the planned intervention,

and subsequently are less likely to have their disorder remediated. By comparison, the low attrition rates evidenced in the current research (8.1% in Study 1 and 5.3% in Study 2) compare favourably to attrition rates reported for other programs (e.g., Trajkovski et al., 2019), and provide support for the acceptable nature of the stuttering management principles implemented.

The clinical validity of the intervention evaluated in this body research is demonstrated in the maintenance and durability of demonstrated gains at three months follow-up. Improvements in fluency were found to be maintained at three months follow up for all three groups of CWS involved in the prospective clinical trial. Positive shifts in child and parent psychosocial measures were also maintained at follow-up for the group of CWS who received the integrated program, with potential long term implications for improved socioemotional wellbeing for both these parents and their children (Bradshaw et al., 2019; Moffitt et al., 2011). The longer-term outcomes for the children receiving the integrated program, and indeed all of the children, have yet to be determined. Future research could employ a longer maintenance period of 12 to 18 months, to ascertain the extent of benefit of this integrated intervention in terms of concurrent ongoing self-regulation and fluency improvements over time.

Parent Reflections

Consumer satisfaction and acceptability are important predictors of engagement with, and success of, health interventions (Sekhon et al., 2017). Therefore, it was deemed important to employ mixed quantitative and qualitative research methodology to allow for exploration of parent experiences with the novel integrated program (Zhang, 2014). In a recent review of mixed methods health intervention research, primary functions of qualitative data were suggested (Zhang, 2014). These include the possibility of using qualitative data to strengthen and inform the design and implementation of an intervention, as well as to explore quantitative data in greater depth. In the current body of research, the primary aim of the qualitative study was to explore the parents' lived experiences and perspectives regarding the integrated intervention overall, and the components therein.

The qualitative study, implemented in Study 3 of this body of research, is the second known published qualitative study to explore parent experiences with early stuttering interventions for their children. A previous study assessed parents' experiences with the Lidcombe program, and shed light on aspects of the Lidcombe

Program that had not yet been recorded in quantitative research (Hayhow, 2009). This provided support for further qualitative investigation relating to parent perceptions and experiences of preschool stuttering intervention.

Major themes that emerged from this preliminary investigation of parent experiences with the program included: the bidirectional relationship between stuttering and child behaviour, improvements in parent and child-self regulation, and general positive perceptions of the program. Findings reinforced the beneficial and acceptable nature of the program expressed by the parents. Such qualitative evidence underscores the importance of tailoring interventions to suit specific profiles of CWS and their families, beyond the quantifiable demonstrated benefit of enhancing stuttering treatment outcomes.

Throughout the parent interviews conducted, parents reflected on the integrated intervention in its entirety, but particularly highlighted the naturalistic and practical elements of the CUSP component of the integrated program. Future research could compare these reflections with those gathered from parents whose children have received different preschool stuttering interventions. This may provide information pertaining to acceptability of different early intervention programs, which could possibly help elucidate the reasons for differing attrition rates across intervention programs (Sekhon et al., 2017).

In line with emerging research investigating optimal methodologies for conducting qualitative interviews with preschool children (aged 3 to 6 years), it is evident that children in this age range are capable of providing highly valuable reflections and insights about their own life experiences (Clark & Statham, 2005; Ponizovsky-Bergelson et al., 2019). Therefore, an additional recommended future research direction would be to implement potential qualitative research conversations with pre-school CWS who have received the CUSP, in order to gain their insights as well.

Theoretical Implications

In this PhD research, each of the three studies undertaken provides support for contemporary multifactorial models of stuttering disorders. These models include the multifactorial dynamic pathways theory (Smith & Weber, 2017), the DCM (Starkweather, 1987) and the dual diathesis stressor framework (Choi et al., 2016; Conture & Walden, 2012; Walden et al., 2012). The multifactorial dynamic pathways theory considers the interplay of vulnerable factors within an individual's

profile that may promote or hinder recovery from stuttering. In considering the emotional factors recognised as central to this theory, it is anticipated that growth in self-regulatory capacities, as experienced by the CWS who received the integrated program, is associated with increased chance of recovery from their stuttering disorder. Authors of the DCM suggest that stuttering is maintained by an imbalance in environmental demands (motoric, linguistic, cognitive and emotional), and the capacity for fluent speech production. By promoting the CWS's capacity to respond to changes in environmental stimuli with less uncontrolled emotional arousal and more desirable behavioural responses through EBPS, excessive emotional demands that impact capacity for fluent speech production is likely to be reduced. Similarly, the dual diathesis stressor framework also encapsulates theoretical support for the concept that emotional reactivity and self-regulation skills are related to the individual's severity of stuttered speech.

In the current body of research, evidence is provided that shows behavioural self-regulation issues may be a contributing factor to the capacity of the child to regulate their fluency. This was demonstrated by the increased stuttering treatment time required for children with behavioural self-regulation issues, when these co-occurring issues were not addressed, compared to CWS without these co-occurring behavioural issues. Furthermore, the clinical trial suggested that when parents promoted development of self-regulation in their children, these skills in turn assisted these children to respond more effectively to stuttering treatment. This resulted in a reduced number of stuttering treatment sessions and a higher likelihood of achieving normal fluency post-treatment and at follow-up, compared to CWS with these behavioural self-regulation issues who did not have them concurrently addressed as part of their stuttering management.

Qualitative comments from parents reflected on the bidirectional relationship between behavioural self-regulation and fluency regulation, and also highlighted the importance of this relationship. For example, one parent reflected that as her child developed the language to label his emotions, it was pivotal for his fluency improvements. Another parent commented that management of child behaviour alongside stuttered speech behaviours worked well together in an integrated way. Both quantitative and qualitative findings provide support for multifactorial models that recognise self-regulation as a factor that may impact the manifestation and severity of developmental stuttering disorders.

Future Considerations

The adapted Level 4 Group Triple P program, an intensive level of Triple P (Sanders et al., 2014), was successful in enhancing stuttering treatment responsiveness and yielding large improvements in child behaviour and parenting outcomes. Implementation of all five levels of Triple P have resulted in significantly improved parenting practices and child behaviours overall, providing support for "lighter touch" or less intensive interventions (Prinz et al., 2009; Sanders et al., 2014). Consequently, CWS who do not present to clinic with challenging behaviour problems as the primary concern (given their primary reason for accessing speech pathology services would likely be for stuttering management), may benefit from a less intensive EBPS. Future research may consider evaluating slightly less-intensive ways of delivering Triple P to test if the benefits for stuttering remains with shorter, less intensive versions of the Triple P program. It is worth noting, however, that the commitment and intervention time dedicated to Triple P was not acknowledged as a limitation in the parent qualitative interviews.

Considering the impact of bias in parent report measures, may further strengthen the internal validity of intervention related changes on measures of child behaviour and parenting practices Although parent report measures used had strong reliability and validity, future studies could consider employing mixed observational and parental report measures of behavioural self-regulation to account for this potential bias. Furthermore, teacher report measures of similar constructs could be considered, in addition to parental report.

A final consideration for future research is optimising allocation to treatment group, while maintaining clinical validity and consumer choice in intervention selection. In the current research, a small proportion of parents self-selected to receive the integrated program despite not being initially assigned to this integrated group (13.8%), while 5.6% elected not to receive the program due to reported parent confidence in management of their child's behaviour. Self-selection in a clinical trial can be viewed as a limitation in terms of reducing randomisation of all participants, and may have contributed to the pre-treatment differences that existed between the eADHD groups on a number of child behaviour and parenting measures. However, the small proportion of self-selection was perceived as a strength in terms of clinical validity. In future research and clinical implementation, a proposed screening measure of parental concern relating to ADHD type symptoms displayed by their

children, may assist in recommendations of the EBPS component to different families.

Application to Other Speech, Language and Communication Disorders

The application and implementation of an adapted or integrated EBPS program for children with a range of speech, language and communication disorders may be beneficial, when the findings applied to this clinical disorder are extrapolated to other speech and language disorders. For example, research pertaining to children with early language difficulties has highlighted a significant proportion of these children to have co-occurring behaviour problems. Similarly to developmental stuttering disorders, the proportion of children with language difficulties who have co-occurring behaviour problems has been reported to be as high as 54%, and the possible impact of these behaviour problems on language intervention outcomes remains to be further investigated (Curtis et al., 2018). The presence of a language disorder has been found to consistently predict childhood behaviour problems (Curtis et al., 2018). As with early stuttering disorders, the parents of children with language difficulties play a significant role in treatment and remediation (Law et al., 2019). Consequently, there may be a similar need to examine the impact of these behaviour problems on treatment outcomes, and potentially to provide support to parents in their management of all aspects of the child's presenting problems in a holistic and family centred manner (Lindsay & Strand, 2016; Ozcebe et al., 2019; Thurm et al., 2018).

Conclusion

This PhD project identified a number of unique and clinically relevant findings relating to the management of early developmental stuttering, particularly for those CWS with co-occurring behavioural self-regulation challenges. Key highlights included the identification of the proportion of CWS who present with concomitant behaviour problems, and the determination of the impact of ADHD symptoms on stuttering treatment responsiveness. In addition, the clinical intervention trial implemented a successful and novel integrated stuttering treatment and self-regulation program, which positively influenced the fluency trajectories for CWS who also had challenging self-regulation issues. This research, therefore, has demonstrated that self-regulation is a particularly important factor to address in developmental stuttering to positively influence stuttering treatment outcomes, for those CWS with weaker self-regulatory capacities. Such findings extend current

theory and contemporary models of early stuttering, by demonstrating how increased self-regulatory capacity may facilitate the pathway to recovery for this subgroup of CWS. Qualitative analyses revealed the novel integrated program to be highly beneficial for the children and their families, and identified the concurrent benefits of supporting parents to manage their child's stuttering difficulties and self-regulation skills simultaneously.

A final important finding is the preliminary evidence for the success of the Curtin University Stuttering Program as a flexible and individualised approach to the management of developmental stuttering disorders. This research has highlighted the importance of the management of early developmental stuttering disorders and self-regulation challenges that may be experienced by the child in their family context.

References

Alm, P. A., & Risberg, J. (2007). Stuttering in adults: The acoustic startle response, temperamental traits, and biological factors. *Journal of Communication Disorders, 40*(1), 1-41. https://doi.org/10.1016/j.jcomdis.2006.04.001.

Ambrose, N., Yairi, E., Loucks, T., Seery, C., & Throneburg, R. (2015). Relation of motor, linguistic, and temperament factors in epidemiologic subtypes of persistent and recovered stuttering: Initial findings. *Journal of Fluency Disorders*. https://doi.org/10.1016/j.fludis.2015.05.004.

American Psychological Association (2013). *American Psychiatric Association: Diagnostic and statistical manual of mental health disorders* (5th ed.). Arlington, VA: American Psychiatric Association.

Anastopoulos, A., & Shelton, T. (2001). *Assessing attention-deficit/hyperactivity disorders*. New York, NY: Kluwer Academic Publishers.

Anderson, J., Pellowski, M., Conture, E., & Kelly, E. (2003). Temperamental characteristics of young children who stutter. *Journal of Speech, Language and Hearing Research, 46*(5), 1221-1233.

Anderson, J., & Wagovich, S. (2010). Relationships among linguistic processing speech, phonological working memory, and attention in children who stutter. *Journal of Fluency Disorders, 35*, 216-234. https://doi.org/10.1016/j.jfludis.2010.04.003.

Anzman-Frasca, S., Francis, L., & Birch, L. (2015). Inhibitory control is associated with psychosocial, cognitive, and weight outcomes in a longitudinal sample of girls. Psychological Science, 1(3), 203–216. https://doi.org/10.1037/tps0000028.

Arnold, H., Conture, E., Key, A., & Walden, T. (2011). Emotional reactivity, regulation and childhood stuttering: A behavioral and electrophysiological study. *Journal of Communication Disorders, 44*(3), 276-293.

Backer-Grøndahl, A., Nærde, A., Ulleberg, P., & Janson, H. (2016). Measuring effortful control using the children's behavior questionnaire-very short form: Modeling matters. *Journal of Personality Assessment, 98*(1), 100–109.

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioral change. *Psychological Review, 84*, 181–215.

Barbour, R. S. (2001). Checklists for improving rigour in qualitative research: a case of the tail wagging the dog? *British Medical Journal, 322*, 1115–1117.

Barkley, R. (2013). *Taking charge of ADHD, third edition: The complete, authoritative guide for parents* (3 ed.). Guildford Publications.

Barros, L., Goes, A. R., & Pereira, A. I. (2015). Parental self-regulation, emotional regulation and temperament: Implications for intervention. *Estudos De Psicologia*, *32*(2), 295–306. https://doi.org/10.1590/0103-166X2015000200013.

Basten, M., Tiemeier, H., Althoff, R., van de Schoot, R., Jaddoe, V., Hofman, A., Hudziak, J., Verhulst, F., & van der Ende, J. (2016). The stability of problem behavior across the preschool years: An empirical approach in the general population. *Journal of Abnormal Child Psychology, 44*, 393-404.

Beilby, J. (2014). Psychosocial impact of living with a stuttering disorder: Knowing is not enough. *Seminars in Speech and Language, 35*(2), 132-143. https://doi.org/10.1055/s-0034-1371756.

Benish, S., Quintana, S., & Wampold, B. (2011). Culturally adapted psychotherapy and the legitimacy of myth: a direct-comparison meta-analysis. *Journal of Counseling Psychology, 58*, 279-289.

Bertakis, K. D., & Azari, R. (2011). Patient-centred care is associated with decreased health care utilization. *Journal of the American Board of Family Medicine*, *24*(3), 229–239.

Binns, A., Hutchison, L., & Oram Cardy, J. (2019). The speech-language pathologist's role in supporting the development of self-regulation: A review and tutorial. *Journal of Communication Disorders, 78*, 1–17. https://doi.org/10.1016/.jcomdis.2019.12.005.

Bjornebekk, G., Kjobli, J., & Ogden, T. (2015). Children with con- duct problems and co-occurring ADHD: Behavioral im- provements following parent managment training. *Child & Family Behavior Therapy,* 37(1), 1–19. https://doi.org/10.1080/ 07317107.2015.1000227

Blair, C., & Diamond, A. (2008). Biological processes in prevention and intervention: The promotion of self-regulation as a means of preventing school failure. *Development and Psychopathology, 20*(3), 889-911. https://doi.org/10.1017/S0954579408000436.

Blair, C., & Ursache, A. (2011). A bidirectional model of executive functions and self-regulation. In K. Vohs & R. Baumeister (Eds.), *Handbook of self-regulation: Research, theory, and applications* (pp. 300-320). Guildford Press.

Blomgren, M. (2013). Behavioral treatments for children and adults who stutter: A review. *Psychology Research and Behavior Management, 6*, 9-19.

Blood, G., Ridenour Jr., V., Qualls, C., & Hammer, C. (2003). Co-occurring disorders in children who stutter. *Journal of Communication Disorders, 36*(6), 427-448. https://doi.org/10.1016/S0021-9924(03)00023-6

Bloodstein, O., & Bernstein Ratner, N. (2008). *A handbook on stuttering* (6 ed.). Thomson Delmar Learning.

Bor, W., Sanders, M., & Markie-Dadds, C. (2002). The effects of the Triple P-Positive Parenting Program on preschool children with co-occurring disruptive behavior and attentional/hyperactive difficulties. *Journal of Abnormal Child Psychology, 30*(6), 571–587. https://doi.org/10.1023/A:1020807613155.

Bradshaw, S., Bem, D., Shaw, K., Taylor, B., Chiswell, C., Salama, M., Bassett, E., Geetinder, K., & Cummins, C. (2019). Improving health, wellbeing and parenting skills in parents of children with special health care needs and medical complexity - a scoping review. *BMC Pediatrics, 19*(301), 1-11.

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77–101. https://doi.org/10.1191/1478088706qp063oa.

Brown, M., McIntyre, L., Crnic, K., Baker, B., & Blacher, J. (2011). Preschool children with and without developmental delay: risk, parenting, and child demandingness. *Journal of Mental Health Research in Intellectual Disabilities, 4*(3), 206–226. https://doi.org/10.1080/19315864.2011.596990.

Campbell, S. M. (2000). *Attention-deficit/hyperactivity disorder: A developmental view.* In A. J. Sameroff, M. Lewis, & S. M. Miller (Eds.), *Handbook of developmental psychopathology* (p. 383–401). Kluwer Academic Publishers.

Carey, B., Onslow, M., & O'Brian, S. (2020a). Natural recovery from stuttering for a clinical cohort of pre-school children who received no treatment. *Journal of Speech-Language Pathology*, 1-9.

Carey, B., Onslow, M., & O'Brian, S. (2020b). Natural recovery from stuttering for a clinical cohort of pre-school children who received no treatment *International Journal of Speech-Language Pathology*. https://doi.org/https://doi.org/10.1080/17549507.2020.1746399.

Carter, A., Breen, L., Yaruss, J. S., & Beilby, J. (2017). Self-efficacy and quality of life in adults who stutter. *Journal of Fluency Disorders, 54*, 14-23. https://doi.org/10.1016/j.fludis.2017.09.004.

Chang, S., Erickson, K., Ambrose, N., Hasegawa-Johnson, M., & Ludlow, C. (2008). Brain anatomy differences in childhood stuttering. *NeuroImage, 39*(3), 1333–1344. https://doi.org/10.1016/j.neuroimage.2007.09.067.

Choi D, Conture E, Walden, T, Jones, R, & Kim, H. (2016). Emotion stress, emotion diathesis and childhood stuttering. J*ournal of Speech, Language, and Hearing Research*, *59*(4), 616-630.

Choi, D., Conture, E., Walden, T., Lambert, W., & Tumanova, V. (2013). Behavioral inhibition in childhood stuttering. *Journal of Fluency Disorders, 38*, 171–183.

Clark, C., Prior, M., & Kinsella, G. (2002). The relationship be- tween executive function abilities, adaptive behavior and academic achievment in children with externalizing behavior problems. *The Journal of Child Psychology and Psychiatr*y, *43*(6), 785–796. https://doi.org/10.1111/1469-7610.00084.

Clark, A., & Statham, C. (2005). Listening to young children: Experts in their own lives. *Adoption and Fostering, 29*, 45-56.

Clark, C., Tumanova, V., & Choi, D. (2017). Evidence-based multifactorial assessment of pre-school age children who stutter. *Perspectives of the ASHA Special Interest Groups, 2*(4), 4–27.

Connery, A., McCurtin, A., & Robinson, K. (2019). The lived experience of stuttering: a synthesis of qualitative studies with implications for rehabilitation. *Disability and Rehabilitation, 29*, 1–11. https://doi.org/10.1080/09638288.2018.1555623.

Conture, E., Kelly, E., & Walden, T. (2013). Temperament, Speech and Language: An Overview. *Journal of Communication Disorders, 46*(2), 125–142. https://doi.org/10.1016/j.jcomdis.2012.11.002.

Conture, E.G. &, Walden, T.A. (2012). *Dual Diathesis-Stressor Model of Stuttering*. In Y.O. Filatova (Ed.), Theoretical Issues of Fluency Disorders (p. 94-127). Moscow: National Book Centre.

Curtis, P., Frey, J., Watson, C., Hampton, L., & Roberts, M. (2018). Language disorders and problem behaviors: A meta-analysis. *Pediatrics, 142*(2), 1-14.

Danforth, J. S., Barkley, R. A., & Stokes, T. F. (1991). Observations of parent-child interactions with hyperactive children: Research and clinical implications. *Journal of the American Academy of Child and Adolescent Psychiatry, 40*, 508–515. https://doi.org/10.1016/0272-7358(91)90127-G.

D'Souza, S., Underwood, L., Peterson, E., Morton, S., & Waldie, K. (2019). Persistence and change in behavioural problems during early childhood. *BMC Pediatrics, 19*(259), 1-10.

De Sonneville-Koedoot, C., Stolk, E., Rietveld, T., & Franken, M.-C. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PLoS One, 10*(7), 1-17. https://doi.org/10.1371/journal.pone.0133758.

Deater-Deckard, K. (2014). Family matters: Intergeneration and interpersonal processes of executive function and attentive behaviour. *Current Directions in Psychological Science, 23*, 230. https://doi.org/10.1177/0963721414531597.

Delaney, L. (2018). Patient-centred care as an approach to improving health care in Australia. *The Australian Journal of Nursing Practice, Scholarship and Research, 25*(1), 119–123. https://doi.org/10.1016/j.colegn.2017.02.005.

Dodd, B., Hua, Z., Crosbie, S., Holm, A., & Ozanne, A. (2002). Diagnostic evaluation of articulation and phonology. The Psychological Corporation.

Donaghy, M., & Smith, K. (2016). Management options for pediatric patients who stutter: current challenges and future directions *Paediatric Health, Medicine and Therapeutics, 7*, 71-77.

Donaher, J., & Richels , C. (2012). Traits of Attention Deficit/Hyperactivity Disorder in school-age children who stutter. *Journal of Fluency Disorders, 37*(4), 242-252. https://doi.org/10.1016/j.jfludis.2012.08.002.

DosReis, S., Barksdale, C., Sherman, A., Maloney, K., & Charach, A. (2010). Stigmatising experiences of parents of children with a new diagnosis of ADHD. *Psychiatric Services, 61*(8), 811–816.

Drayna, D., & Frigerios-Domingues, C. (2017). Genetic contributions to stuttering: the current evidence. *Molecular Genetics and Genomic Medicine, 5*(2), 95-102.

Druker, K., Mazzucchelli, T. G., & Beilby, J. (2019). An evaluation of integrated fluency and resilience program for early developmental stuttering disorders. *Journal of Communication Disorders, 78*, 69-83. https://doi.org/10.1016/j.jcomdis.2019.02.002.

Duckworth, A. L., & Seligman, M. (2005). Self-discipline outdoes IQ in predicting academic performance in adolescents. Psychological Science, *16*(12), 939–944. https://doi.org/10.1111/j.1467- 9280.2005.01641.x

DuPaul, G. J., McGoey, K. E., Eckert, T. L., & VanBrakel, J. (2001). Preschool children with attention-deficit/hyperactivity disorder: Impairments in behavioural, social and school functioning. *Journal of American Academy of Child and Adolescent Psychiatry, 40*, 508–515. https://doi.org/10.1097/00004583-200105000-00009.

DuPaul, G. J., Anastopoulos, A. D., Power, T. J., Reid, R., Ikeda, M. J., & McGoey, K. E. (1998). Parent ratings of attention- deficit/hyperactivity disorder symptoms: Factor structure and normative data. J*ournal of Psychopathology and Behavioral Assessment*, *20*, 83–102. https://doi.org/10.1023/A:1023087410712.

Easterbrook, C. J., & Meehan, T. (2017). The therapeutic relationship and cognitive behavioural therapy: A case study of an adolescent girl with depression. *The European Journal of Counselling Psychology, 6*(1), 1–24. https://doi.org/10.5964/ejcop.v6i1.85.

Eggers, K., De Nil, L., & Van den Bergh, B. (2010). Temperament dimensions in stuttering and typically developing children. *Journal of Fluency Disorders, 35*, 355-372. https://doi.org/10.1016/j.jfludis.2010.10.004.

Eggers, K., De Nil, L., & Van Den Bergh, B. (2013). Inhibitory control in childhood stuttering *Journal of Fluency Disorders, 38*, 1-13. https://doi.org/10.1016/j.jfludis.2012.10.001

Eggers, K., De Nil, L. F., & Van Den Bergh, B. R. (2012). The efficiency of attentional networks in children who stutter. *Journal of Speech, Language and Hearing Research, 55*(3), 946-959. https://doi.org/10.1044/1092-4388(2011/10-0208).

Eisele, M., Winfried, A., Rakebrandt, A., Boczor, S., Blozik, E., Trader, J., Stork, S., Herrmann-Lingen, C., & Scherer, M. (2018). Importance of comorbidities in the treatment of primary care patients with heart failure - baseline results of the observational RECODE-HF Study. *Family Practice 35*(4), 481-487, https://doi.org/10.1093/fampra/cmx135

Eisenberg, N., & Spinrad, T. (2004). Emotion-related regulation: Sharpening the definition. *Child Development*, *75*(2), 334–339 https://doi.org/10.1111/j.1467-8624.2004.00674.x

Eisenberg, N., & Sulik, M. J. (2012). Emotion-related self-regulation in children. *Teaching of Psychology, 39*(1), 77–83. https://doi.org/10.1177/0098628311430172.

Eisenberg, N., Zhou, Q., Spinrad, T., Valiente, C., Fabes, R., & Liew, J. (2005). Relations among positive parenting, children's effortful control, and externalizing problems: A three-wave longitudinal study. *Journal of Child Development,76*(5), 1055–1071. https://doi.org/10.1111/j.1467-8624.2005.00897.x.

Ekman, I., Wolf, A., Olsson, L., Taft, C., Dudas, K., Schaufelberger, M., ... Swedberg, K. (2012). Effects of person-centred care in patients with chronic heart failure: The PCC-HF study. *European Heart Journal, 33*(9), 1112–1119.

Ellis, B., & Nigg, J. (2009). Parenting practices and attention-deficit/hyperactivity disorder: new findings suggest partial specificity of effects. *Journal of American Academy of Child and Adolescent Psyhchiatry, 48*(2), 146–154. https://doi.org/10.1097/CHI.0b013e31819176d0.

Embrechts, M., Ebben, H., Franke, P., & Van de Poel, C. (2000). Temperament: A comparison between children who stutter and children who do not stutter. Third World Congress on fluency disorders, Nijmegen, The Netherlands: University of Nijmegen Press.

Eyberg, S. (1988). Parent-child interaction therapy: Integration of traditional and behavioral concerns. *Child and Behavior Therapy, 10*, 33–46. https://doi.org/10. 1300/J019v10n01_04.

Fagnani, C., Fibiger, S., Skythe, A., & Hjelmborg, J. (2011). Heritability and environmental effects for self-reported periods with stuttering: a twin study from Denmark. *Logopedics Phoniatrics Vocology, 36*(3), 114-120.

Felsenfeld, S., Van Beljsterveldt, C., & Boomsma, D. I. (2010). Attentional regulation in young twins with probable stuttering, high nonfluency, and typical fluency. *Journal of Speech, Language, and Hearing Research, 53*(5), 1147-1166. https://doi.org/10.1044/1092-4388(2010/09-0164.

ForFluency (2012). Fluency Rater. [Mobile application software]. Retrieved from http://download.cnet.com/Fluency-Rater/3000-2129_4-75357634.html.

Francis, L., & Susman, E. (2009). Self-regulation and rapid weight gain in children from age 3 to 12 years. *Archives of Pediatrics & Adolescent Medicine*, *163*(4), 297–302. https://doi.org/10.1001/ archpediatrics.2008.579

Franken, M. C., & Putker-de Bruijn, D. (2007). Restart-DCM Method. *Treatment protocol developed within the scope of the ZonMW project: Cost Effectiveness of the Demands and Capacities Model based treatment compared to the Lidcombe program of early stuttering: Randomised trial.* *http://www.nedverstottertherapie.nl*.

Fujiwara, T., Kato, N., & Sanders, M. (2011). Effectiveness of Group Positve Parenting Program (Triple P) in changing child behavior, parenting style, and parental adjustment: An intervention study in Japan. *Journal of Child and Family Studies, 20*(6), 804–813. https://doi.org/10.1007/s10826-011-9448-1

Goodman, A., & Goodman, R. (2012). Strengths and Difficulties Questionnaire scores and mental health in looked after children. *The British Journal of Psychiatry*, *200*(5), 426–427. https://doi. org/10.1192/bjp.bp.111.104380

Goodman, R. (1997). The Strengths and Difficulties Questionnaire: A research note. *The Journal of Child Psychology and Psychiatry*, *38*, 581–586. https://doi.org/10.1111/j.1469-7610.1997. tb01545.x

Goodman, R., Ford, T., Simmons, H., Gatward, R., & Meltzer, H. (2000). Using the Strengths and Difficulties Questionnaire (SDQ) to screen for child psychiatric disorders in a community sample. *British Journal of Psychiatry*, *117*(6), 534–539. https://doi.org/ 10.1192/bjp.177.6.534

Goodman, R., Renfrew, D., & Mullick, M. (2000). Predicting type of psychiatric disorder from Strengths and Difficulties Ques- tionnaire (SDQ) scores in child mental health clinics in London and Dhaka. *European Child & Adolescent Psychiatry*, *9*, 129–134. https://doi.org/10.1007/s007870050008

Gray, G., Totsika, V., & Lindsay, G. (2018). Sustained effectiveness of evidence-based parenting programs after the research trial ends. *Frontiers in Psychology, 9*, 1-11.

Grey, M., Goodrich, A., Jaser, S., & Lindemann, E. (2011). Coping skills traning for parents of children with Type 1 diabetes: 12-month outcomes. *Nursing Research*, *60*(3), 173–181. https:// doi.org/10.1097/NNR.0b013e3182159c8f

Gross, H., Shaw, D., & Moilanen, K. (2008). Reciprocal associations between boys' externalizing problems and mothers' depressive symptoms. *Journal of Abnormal Child Psychology, 36*, 693–709. https://doi.org/10.1007/s10802-008-9224.

Guest, G., Bunce, A., & Johnson, L. (2006). How many interviews are enough? An experiment with data saturation and variability. *Field Methods, 18*(1), 59–82. https:// doi.org/10.1177/1525822X05279903.

Guitar, B. (2014). *Stuttering: An integrated approach to its nature and treatment* (4th ed.). Philadelphia, PA: Lippincott Williams & Wilkins.

Guitar, B., Kazenski, D., Howard, A., Cousins, S., Fader, E., & Haskell, P. (2015). Predicting treatment time and long-term outcomes of the Lidcombe Program: A replication and reanalysis. *American Journal of Speech-language Pathology, 24*(3), 533–544.

Haskett, M. E., Armstrong, J., Neal, S., & Aldianto, K. (2018). Perceptions of Triple P-Positive Parenting Program seminars among parents experiencing homelessness. *Journal of Child and Family Studies, 27*, 1957–1967. https://doi.org/10.1007/s10826-018-1016-5.

Hawes, D., & Dadds, M. (2006). Assessing parenting practices through parent report and direct observation during parent training. *Journal of Child and Family Studies*, *15*, 554–567. https://doi.org/10.1007/s10826-006-9029-x

Hayhow, R. (2009). Parents' experiences of the Lidcombe Program of early stuttering intervention. *International Journal of Speech-Language Pathology, 11*(1), 20-25.

Hoath, F., & Sanders, M. (2002). A feasibility study of Enhanced Group Triple P - positive parenting program for parents of children with Attention-deficit/hyperactivity Disorder. *Behaviour Change, 19*(4), 191-206. http://catalogue.curtin.edu.au/primo_library/.

Houlding, C., Schmidt, F., Stern, S. B., Jamieson, J., & Borg, D. (2012). The perceived impact and acceptability of Group Triple P Positive Parenting Program for Aboriginal parents in Canada. *Children and Youth Services Review, 34*(12), 2287–2294. https://doi.org/10.1016/j.childyouth.2012.08.001.

Howell, P., Davis, S., Patel, H., Cuniffe, P., Downing-Wilson, D., Au-Yeung, J., & Williams, R. (2004). Fluency development and temperament in fluent children and children who do not stutter. In *Theory, research and therapy in fluency disorders. Proceeding of the fourth world congress on fluency disorders.* (pp. 250-256). The Netherlands: University Press Nijmegen.

Huguet, A., Eguren, J., Miguel-Ruiz, D., Valles, X., & Alda, A. (2019). Deficient emotional self-regulation in children with attention deficit hyperactivity disorder: Mindfulness as a useful treatment modality. *Journal of Developmental and Behavioral Pediatrics, 40*(6), 425-431.

Jones, R., Buhr, A., Conture, E., Tumanova, V., Walden, T., & Porges, S. (2014a). Autonomic nervous system activity of preschool-age children who stutter. *Journal of Fluency Disorders, 41*, 12-31.

Jones, R., Choi, D., Conture, E., & Walden, T. (2014b). Temperament, emotion and childhood stuttering. *Seminars in Speech and Language, 35*(2), 114-131. https://doi.org/10.1055/s-0034-1371755.

Jones, R., Conture, E., & Walden, T. A. (2014c). Emotional reactivity and regulation associated with fluent and stuttered utterances of preschool-age children who stutter. *Journal of Communication Disorders, 48*, 38-51. https://doi.org/10.1016/j.jcomdis.2014.02.001.

Kain, M., Bolker, B., & McCoy, M. (2015). A practical guide and power analysis for GLMMs: Detecting among treatment variation in random effects. *PeerJ, 3*(1226), 1–24.

Karrass, J., Walden, T., Conture, E., Graham, C., Arnold, H., Hartfield, K., & Schwenk, K. (2006). Relation of emotional reactivity and regulation to childhood stuttering. *Journal of Communication Disorders, 39*( ), 402-423. https://doi.org/10.1016/j.jcomdis.2005.12.004.

Karimi, H., Jones, M., O'Brian, S., & Onslow, M. (2014). Clinician percent syllables stuttered, clinician severity ratings and speaker severity ratings: Are they

interchangeable? *International Journal of Language & Communication Disorders*, *49*(3), 364–368.
https://doi.org/10.1111/1460-6984.12069

Kefalianos, E., Onslow, M., Ukoumunne, O., Block, S., & Reilly, S. (2014). Stuttering, temperament, and anxiety: Data from a community cohort ages 2-4 years. Journal of Speech, Language, and Hearing Research. *37*(3), 151–163.

Kernick, D. (2003). Introduction to health economics for the medical practitioner. *Postgraduate Medical Journal*, *79*, 147–150.
https://doi.org/10.1136/pmj.79.929.147.

King, K., Alexander, D., & Seabi, J. (2016). Siblings' perceptions of their ADHD-diagnosed sibling's impact on the family system. *International Journal of Environmental Research and Public Health, 13*, 910–920.
https://doi.org/10.3390/ijerph13090910.

Kingston, M., Huber, A., Onslow, M., Jones, M., & Packman, A. (2003). Predicting treatment time with the Lidcombe Program: replication and meta-analysis. *International Journal of Language and Communication Disorders, 38*(2), 165-177.

Klein, J., & Hood, S. (2004). The impact of stuttering on employment opportunities and job performance. *Journal of Fluency Disorders, 29*, 255-273.

Klompas, M., & Ross, E. (2004). Life experiences of people who stutter, and the perceived impact of stuttering on quality of life: personal accounts of South African individuals. *Journal of Fluency Disorders, 29*, 275-305.

Kraft, S. J., Ambrose, N., & Chon, H. (2014). Temperament and environmental contributions to stuttering severity in children: the role of effortful control. *Seminars in Speech and Language, 35*(2), 80-94. https://doi.org/10.1055/s-0034-1371753.

Kraft, S. J., Beilby, J., & Lowther, E. (2019). The role of effortful control in stuttering severity in children: Australian replication study. *American Journal of Speech-Language Pathology*, 1-15. https://doi.org/10.1044/2018_AJSLP-17-0097.

Langevin, M., Packman, A., & Onslow, M. (2009). Peer responses to stuttering in the preschool setting. *American Journal of Speech-Language Pathology, 18*, 264-276.

Langevin, M., Packman, A., & Onslow, M. (2010). Parent perceptions of the impact of stuttering on their preschoolers and themselves. *Journal of Communication Disorders, 43*(5), 407-423. https://doi.org/10.1016/j.jcomdis.2010.05.003.

Law, J., Levickis, P., Rodriguez-Ortiz, I., Matic, A., Lyons, R., Messarra, C., Hreich, E., & Stankova, M. (2019). Working with the parents and families of children with developmental language disorders: An international perspective. *Journal of Communication Disorders, 82*, 1-12.

Lifford, K., Harold, G., & Thapar, A. (2008). Parent-child relationships and ADHD symptoms: A longitudinal analysis *Journal of Abnormal Child Psychology, 36*(2), 285-296. https://doi.org/10.1007/s10802-007-9177-5.

Lindsay, G., & Strand, S. (2016). Children with language impairment: Prevalence, associated difficulties, and ethnic disproportionality in an English population. *Frontiers in Education, 1*, 1-14.

MacPherson, M., & Smith, A. (2013). Influences of sentence length and syntactic complexity on the speech motor control of children who stutter. *Journal of Speech, Language and Hearing Research, 56*, 89-102. https://doi.org/10.1044/1092-4388(2012/11-0184).

Manning, W. (2010). *Clinical decision making in fluency disorders.* San Diego, CA: Singular/Thomson Learning.

Manning, W. H., & DiLollo, A. (2018). *Clinical decision making in fluency disorders*. Plural Publishing Incorporated.

Markie-Dadds, C., Turner, K., & Sanders, M. (2015). *Every parent's group workbook: Triple P positive parenting solutions* (3rd ed.). Triple P International.

Mazzucchelli, T., & Sanders, M. (2018). Children with externalizing behavior problems. In M. Sanders & T. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system* (pp. 85–98). Oxford University Press.

Mazzucchelli, T., G. (2018). Social learning influences: modelling, instructions, consequences. In M. Sanders & A. Morawska (Eds.), *Handbook of Parenting and Child Development Across the Lifespan* (pp. 67-96). Springer International Publishing.

Mazzucchelli, T. G., & Sanders, M. R. (2010). Facilitating practi- tioner flexibility within an empirically supported intervention: Lessons from a system of

parenting support. Clinical Psychology Science and Practice, 17(3), 238–252. https://doi.org/10.1111/ j.1468-2850.2010.01215.x

McAllister, J. (2016). Behavioural, emotional and social development of children who stutter. *Journal of Fluency Disorders, 50*, 23-32. https://doi.org/10.1016/j.fludis.2016.09.003.

McCulloch, J., Swift, M., & Wagnitz, B. (2017). Case file audit of Lidcombe program outcomes in a student-led stuttering clinic. *International Journal of Speech-Language Pathology, 19*(2), 165-173. https://doi.org/10.3109/1754-9515.

McGoey, K., DuPaul, G., Haley, E., & Shelton, T. (2007). Parent and teacher ratings of attention-deficit/hyperactivity disorder in preschool: The ADHD rating scale-IV preschool version. *Journal of Psychopathology and Behavioral Assessment, 29*, 269–276.

Mikami, A. Y., & Pfiffner, L. J. (2008). Sibling relationships among children with ADHD. *Journal of Attention Disorders, 11*, 482-492. https://doi.org/10.1177/1087054706295670.

Millard, S., Nicholas, A., & Cook, F. (2008). Is parent-child interaction therapy effective in reducing stuttering? *Journal of Speech, Language and Hearing Research, 51*(3), 636-650.

Millard, S. K., & Davis, S. (2016). The Palin Parent Rating Scales: Parents' perspectives of childhood stuttering and its impact *Journal of Speech, Language and Hearing Research, 59*, 950-963. https://doi.org/10.1044/2016_JSLHR-S-14-0137.

Millard, S. K., Zebrowski, P., & Kelman, E. (2018). Palin Parent-Child Interaction Therapy: the bigger picture. *American Journal of Speech-Language Pathology, 27*, 1211-1223. https://doi.org/10.1044/2018_AJSLP-ODC11-17-0199.

Miller, A., Gouley, K., Seifer, R., Dickstein, S., & Shields, A. (2004). Emotions and behavior in the Head Start classroom: Associations among observed dysregulation, social competence, and preschool adjustment. *Early Education and Development, 15*, 147-165.

Minnes, P., Perry, A., & Weiss, J. (2015). Predictors of distress and well-being in parents of young children with developmental disabilities: the importance of

parent perceptions. *Journal of Intellectual Disability Research*, *59*(6), 551–
560. https://doi.org/10.1111/jir.12160.

Moffitt, T., Arseneault, L., Belsky, D., Dickson, N., Hancox, R. J., Harrington, H.,
Houts, R., Poulton, R., Roberts, B. W., Ross, S., Sears, M. R., Thomson, W.
M., & Caspi, A. (2011). Gradient of childhood self-control predicts health,
wealth, and public safety. *Proceedings of the National Academy of Sciences
of the United States of America, 108*(7), 2693-2698.
https://doi.org/10.1073/pnas.1010076108

Neece, C., Green, S., & Baker, B. (2012). Parenting stress and child behavior
problems: A transactional relationship across time. *American Journal on
Intellectual and Developmental Disabilities, 117*, 48–66.
https://doi.org/10.1352/1944-7558-117.1.48.

Norris, L. (2016). *Self-regulation strategies for students with disruptive behavior
disorders* [Master's thesis, St. Cloud State University]. theRepository at St.
Cloud State: Culminating Projects in Special Education.

NSCAW. (2009). *Inhibitory control abilities among young children in the child
welfare system.* U.S. Department of Health & Human Services,
Administration for Children & Families: National Survey of Child and
Adolescent Well-Being.

Ntourou, K., Anderson, J., & Wagovich, S. (2018). Executive function and childhood
stuttering: Parent ratings and evidence from a behavioral task. *Journal of
Fluency Disorders*, 56, 18–32. https:// doi.org/10.1016/j.jfludis.2017.12.001

Ntourou, K., Conture, E., & Walden, T. (2013). Emotional reactivity and regulation
in preschool-age children who stutter. *Journal of Fluency Disorders, 38*, 260-
274.

Nye, C., Vanryckeghem, M., Schwartz, J., Herder, C., Turner, H., & Howard, C.
(2013). Behavioral stuttering interventions for children and adolescents: A
systematic review and meta-analysis. *Journal of Speech, Language and
Hearing Research, 56*, 921-932.

O'Brian, S., Jones, M., Packman, A., Menzies, R., & Onslow, M. (2011). Stuttering
severity and educational attainment. *Journal of Fluency Disorders, 36*, 86-92.
https://doi.org/10.1016/j.jfludis.2011.02.006.

Oberklaid, F. (2014). Prevention and early detection in young chil- dren: Challenges for policy and practice. The Medical Journal of Australia, 201(7), 369–370. https://doi.org/10.5694/mja14.01200.

Onchwari, G., & Keengwe, J. (2011). Examining the relationship of children's behavior to emotion regulation ability. *Early Childhood Education Journal, 39*, 279–284.

Onslow, M., & O'Brian, S. (2013). Management of childhood stuttering. *Journal of Paediatrics and Child Health, 49*(2), 112-115. https://doi.org/10.1111/jpc.12034.

Onslow, M., Packman, A., & Harrison, E. (2003). *The Lidcombe Program of early stuttering intervention: A clinician's guide* Pro-Ed.

Ozcebe, E., Erbas, A., & Tigrak, T. (2019). Analysis of behavioural characteristics of children with developmental language disorders. *International Journal of Speech-Language Pathology*.

Palinkas, L., Aarons, G., & Horwitz, S. (2011). Mixed methods designs in implementation research. *Administration and Policy in Mental Health and Mental Health Services Research, 38*(1), 44-53.

Patterson, G. R. (2005). The next generation of PMTO models. *Behavioral Therapist, 28*(2), 27–33.

Peters, K., & Jackson, D. (2008). Mothers' experiences of parenting a child with attention deficit hyperactivity disorder. *Journal of Advanced Nursing, 65*, 62–71. https://doi.org/10.1111/j.1365-2648.2008.04853.x.

Petrovic, P., & Castellanos, F. (2016). Top-down regulation from ADHD to emotional instability. Frontiers in *Behavioral Neuroscience, 10*, 70. https://doi.org/10.3389/fnbeh.2016.00070.

Pidano, A., & Allen, A. (2015). The Incredible Years series: A review of the independant research base. *Journal of Child and Family Studies, 24*, 1898-1916.

Plankers, T. (1999). Speaking in the claustrum: The psychodynamics of stuttering. *International Journal of Psycho-Analysis, 80*(2), 239-256.

Plexico, L., & Burrus, E. (2012). Coping with a child who stutters: a phenomenological analysis. *Journal of Fluency Disorders, 37*, 275-288. https://doi.org/10.1016/j.jfludis.2012.06.002.

Ponizovsky-Bergelson, Y., Dayan, Y., Wahle, N., & Roer-Strier, D. (2019). A qualitative interview with young children: What encourages or inhibits young children's participation? *International Journal of Qualitative Methods, 18*, 1-9.

Posner, M. I., & Rothbart, M. K. (2009). Toward a physical basis of attention and self-regulation. *Physics of Life Review, 6*(2). https://doi.org/10.1016/j.plrev.2009.02.001.

Power, T. J., Costigan, T. E., Leff, S. S., Eiraldi, R. B., & Landau, S. (2001). Assessing ADHD across settings: Contributions of behavioral assessment to categorical decision making *Journal of Clinical Child & Adolescent Psychology, 30*(3), 399-412. https://doi.org/10.1207/S15374424JCCP3003_11.

Prinz, R. J., Sanders, M. R., Shapiro, C. J., Whitaker, D. J., & Lutzker, J. R. (2009). Population-based prevention of child maltreatment: The U.S. Triple P system population trial. *Prevention Science, 10*(1), 1-12.

Reid, R., Trout, A., & Schartz, M. (2005). Self-regulation interventions for children with attention deficit/hyperactivity disorder. *Exceptional Children, 71*, 361-377.

Reilly, S., Onslow, M., Packman, A., Cini, E., Conway, L., Ukoumunne, O., Bavin, E., Prior, M., Eadie, P., Block, S., & Wake, M. (2013). Natural history of stuttering to 4 years of age: a prospective community-based study. *Pediatrics, 132*(3), 460-467. https://doi.org/10.1542/peds.2012-3067.

Riley, G., & Riley, J. (2000). A revised component model for diagnosing and treating children who stutter *Contmeporary Issues in Communication Sciences and Disorders, 27*, 188-199 http://catalogue.curtin.edu.au/primo_library/

Rothbart, K., Ahadi, S., Hershey, K., & Fisher, P. (2001). Investigations of temperament at three to seven years: The Children's Behavior Questionnaire. *Child Development, 72*(5), 1394-1408. https://doi.org/10.1111/1467-8624.00355.

Rothbart, K., Ellis, L., Rueda, R., & Posner, M. (2003). Developing mechanisms of temperament control. *Journal of Personality, 71*(6), 1113-1143.

Rothbart, K., Sheese, B., Rueda, R., & Posner, M. (2011). Devel- oping mechanisms of self-regulation in early life. *Emotion Review*, *3*(2), 207–213. https://doi.org/10.1177/1754073910387943

Rousseau, I., Packman, A., Onslow, M., Harrison, E., & Jones, M. (2007). An investigation of language and phonological development and the responsiveness of preschool age children to the Lidcombe program. *Journal of Communication Disorders, 40*(5), 382–397.

Rueda, R., Posner, M., & Rothbart, K. (2005). The development of executive attention: Contribution to the emergence of self regulation. *Developmental Neuropsychology, 28*, 573–594.

Sameroff, A. (2010). A unified theory of development: A dialectic integration of nature and nurture. *Child Development, 81*(1), 6-22. https://doi.org/10.1111/j.1467-8624.2009.01378.x.

Samyn, V., Roeyers, H., & Bijttebier, P. (2011). Effortful control in typically developing boys and in boys with ADHD or autism spectrum disorder. *Research in Developmental Disabilities, 32*, 483–490. https://doi.org/10.1016/j.ridd.2010.12.038.

Sanchez, M., Lavigne, R., Romero, J., & Elosegui, E. (2019). Emotion regulation in participants diagnosed with attention deficit hyperactivity disorder, before and after an emotion regulation intervention. *Frontiers in Psychology*, 10, 1092. https://doi.org/ 10.3389/fpsyg.2019.01092

Sanders, M. (2003). Triple P – Positive Parenting Program: A population approach to promoting competent parenting. A*ustralian e-Journal for the Advancement of Mental Health*, *2*(3), 127–143. https://doi.org/10.5172/jamh.2.3.127.

Sanders, M. (2012). Development, evaluation, and multinational dissemination of the Triple P-Positive Parenting Program. *Annual Review of Clinical Psychology, 8*, 345-379. https://doi.org/10.1146/annurev-clinpsy-032511-143104.

Sanders, M., Kirby, J., Tellegen, C., & Day, J. (2014). The Triple P-Positive Parenting Program: A systematic review and meta-analysis of a multi-level system of parenting support. *Clinical Psychology Review, 34*(4), 337-357. https://doi.org/10.1016/j.cpr.2014.04.003.

Sanders, M. R., & Mazzucchelli, T., G. (2013). The promotion of self-regulation through parenting interventions. *Clinical Child and Family Psychology Review, 16*(1), 1-17. https://doi.org/10.1007/s10567-013-0129-z.

Sanders, M. R., & Mazzucchelli, T. G. (2018). *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system*. Oxford University Press.

Sanders, M. R., Morawska, A., Haslam, D. M., Filus, A., & Fletcher, R. (2014). Parenting and Family Adjustment Scales (PAFAS): Validation of a brief parent-report measure for use in assessment of parenting skills and family relationships. *Child Psychiatry & Human Development*, *45*(3), 255–272. https://doi. org/10.1007/s10578-013-0397-3

Schwenk, K., Conture, E., & Walden, T. (2007). Reaction to background stimulation of preschool children who do and do not stutter. *Journal of Communication Disorders, 40*(2), 128-441. https://doi.org/10.1016/j.jcomdis.2006.06.003.

Sekhon, M., Cartwright, M., & Francis, J. (2017). Acceptability of healthcare interventions: An overview of reviews and development of a theoretical framework *BMC Health Services Research, 17*(88), 1-13.

Shafiei, B., Faramarzi, S., Abedi, A., Dehqan, A., & Scherer, R. (2019). Effects of the Lidcombe Program and Parent-Child Interaction Therapy on stuttering reduction in preschool children. *Folia Phoniatrica et Logopaedica*, *71*, 29–41. https://doi.org/10.1159/000493915.

Shaw, D. S., Owens, E. B., Giovannelli, J., & Winslow, E. (2001). Infant and toddler pathways leading to early externalising disorders. *Journal of the American Academy of Child & Adolescent Psychiatry*, *40*(1), 36–43. https://doi.org/10.1097/00004583- 200101000-00014

Sheppes, G., Suri, G., & Gross, J. (2015). Emotion regulation and psychopathology. *The Annual Review of Clinical Psychology, 11*, 379-405. https://doi.org/10.1146/annurev-clinspy-032814-112379.

Sidavi, A., & Fabus, R. (2010). A review of stuttering intervention approaches for preschool-age and elementary school-age children. *Contemporary Issues in Communication Sciences and Disorders, 37*, 14-26.

Smith, A. (1999). Stuttering: A unified approach to a multifactorial, dynamic disorder. In *Stuttering research and practice: Bridging the gap* (pp. 27-44). Psychology Press.

Smith, A., & Weber, C. (2016). Childhood stuttering: Where are we and where are we going? *Seminars in Speech and Language*, *37*(4), 291–297. https://doi.org/10.1055/s-0036-1587703.

Smith, A., & Weber, C. (2017). How stuttering develops: the multifactorial dynamic systems pathways theory. *Journal of Speech, Language and Hearing Research, 60*, 2483-2505. https://doi.org/10.1044/2017_JSLHR-S-16-0343.

Smith-Donald, R., Raver, C., Hayes, T., & Richardson, B. (2007). Preliminary construct and concurrent validity of the Preschool Self-regulation Assessment (PSRA) for field-based research. *Early Childhood Research Quarterly*, *22*(2), 173–187. https://doi.org/10.1016/j.ecresq.2007.01.002

Sonney, J., & Insel, K. (2016). Reformulating the common sense model of self-regulation: toward parent-child shared regulation. *Nursing Science Quarterly*, *29*(2), 154–159. https://doi.org/10.1177/0894318416630091.

Sonuga-Barke, E. J. J., Daley, D., Thompson, M., Laver-Bradbury, C., & Weeks, A. (2001). Parent-based therapies for preschool attention-deficit/hyperactivity disorder: A randomised, controlled trial with a community sample. *Journal of American Academt of Child and Adolescent Psyhciatry*, *40*, 402–408. https://doi.org/10.1097/ 00004583-200104000-00008.

Spencer, C., & Weber-Fox, C. (2014). Preschool speech articulation and nonword repetition abilities may help predict eventual recovery or persistence of stuttering. *Journal of Fluency Disorders, 41*, 32-46. https://doi.org/10.1016/j.jfludis.2014.06.001.

Spencer, T., Faraone, S., Surman, C., Petty, C., Clarke, A., Batchelder, H., Wozniak, J., & Biederman, J. (2011). Towards defining deficient emotional self regulation in youth with attention deficit hyperactivity disorder using the child behavior checklist. *Postgraduate Medical Journal, 123*(5), 50-59.

Stafford, M., Kuh, D. L., Gale, C. R., Mishra, G., & Richards, M. (2016). Parent-child relationships and offspring's positive mental wellbeing from adolescence to early older age. *The Journal of Positive Psychology*, *11*(3), 326–337. https://doi.org/10.1080/17439760.2015.1081971.

Starkweather, W. C. (1987). *Fluency and stuttering*. Prentice-Hall.

Starkweather, W. C. (2002). The epigensis of stuttering. *Journal of Fluency Disorders, 27*(4), 269-287. Retrieved from www.sciencedirect.com

Starkweather, W. C., & Ridener Gottwald, S. (1990). The Demands and Capacities Model II: Clinical applications. *Journal of Fluency Disorders, 15*, 143-157.

Starkweather, W. C., Ridener Gottwald, S., & Halfond, M. (1997, 11.3.1997). *Stuttering prevention: a manual for parents*. Temple University Stuttering Prevention Clinic. Retrieved 12.1 from https://www.mnsu.edu/comdis/kuster/Parents/starkweather.html

Thurm, A., Manwaring, S., Jimenez, C., Swineford, L., Farmer, C., Gallo, R., & Maeda, M. (2018). Social-emotional and behavioral problems in toddlers with language delay. *Infant Mental Health Journal, 39*(5), 569-580.

Throneburg, R., & Yairi, E. (2001). Durational, proportionate and absolute frequency characteristics of dysfluencies: A longitudinal study regarding persistence and recovery. *Journal of Speech Language and Hearing Research, 44*(38–51).

Tominey, S., & McClelland, M. (2011). Red light, purple light: Findings from a randomized trial using circle time games to improve behavioral self-regulation in preschool. *Early Education and Development*, *22*(3), 489–519. https://doi.org/10.1080/10409289.2011.574258

Trajkovski, N., O'Brian, S., Onslow, M., Packman, A., Lowe, R., Menzies, R., Jones, M., & Reilly, S. (2019). A three-arm randomized controlled trial of Lidcombe Program and Westmead Program for early stuttering interventions. *Journal of Fluency Disorders, 61*.

Tsukayama, E., Toomey, S. L., Faith, M. S., & Duckworth, A. L. (2010). Self-control as a protective factor against overweight status in the transition from childhood to adolescense. *Archives of Pediatrics and Adolescent Medicine, 164*, 631-635. https://doi.org/10.1001/archpedriatics.2010.97.

Ullebø, A., Posserud, M.-B., Heiervang, E., Gillberg, C., & Obel, C. (2011). Screening for the attention deficit hyperactivity disorder phenotype using the strength and difficulties questionnaire. *European Child & Adolescent Psychiatry*, *20*(9), 451–458. https:// doi.org/10.1007/s00787-011-0198-9.

Ullsperger, J., Nigg, J., & Nikolas, M. (2016). Does child temperament play a role in the association between parenting practices and child attention deficit/hyperactivity disorder? *Journal of Abnormal Child Psychology, 44*, 167-178. https://doi.org/10.1007/s10802-015-9982-1.

Van Borsel, J., Brepoels, M., & De Coene, J. (2011). Stutteirng, attractiveness and romantic relationships: The perception of adolescents and young adults. *Journal of Fluency Disorders, 36*, 41-50.

Van Eerdenbrugh, S., Packman, A., O'Brian, S., & Onslow, M. (2018). Challenges and strategies for speech-language pathologists using the Lidcombe program for early stuttering. *American Journal of Speech-Language Pathology*, *27*, 1259–1272. https://doi.org/10.1044/2014_JSLHR-S-13-0090.

Van Steijn, D., Oerlemans, A., van Aken, M., Buitelaar, J., & Rommelse, N. (2014). The reciprocal relationship of ASD, ADHD, depressive symptoms and stress in parents of children with ASD and/or ADHD. *Journal of Autism and Developmental Disorders, 44*, 1064-1076. https://doi.org/10.1007/s10803-013-1958-9.

Vanryckeghem, M., Brutten, G. J., & Hernandez, L. M. (2005). A comparative investigation of the speech-associated attitude of preschool and kindergarten children who do and do not stutter. *Journal of Fluency Disorders, 30*(4), 307-318. https://doi.org/10.1016/j.jfludis.2005.09.003.

Walden, T., Frankel, C., Buhr, A., Johnson, K., Conture, E., & Karrass, J. (2012). Dual diathesis-stressor model of emotional and linguistic contributions to developmental stuttering. *Journal of Abnormal Child Psychology, 40*(4), 633-644. https://doi.org/10.1007/s10802-011-9581-8.

Walsh, B., Mettel, K., & Smith, A. (2015). Speech motor planning and execution deficits in early childhood stuttering. *Journal of Neurodevelopmental Disorders, 7*, 27. https://doi.org/10.1186/s11689-015-9123-8.

Wampold, B. (2015). How important are the common factors in psychotherapy? An update. *World Psychiatry: The World Psychiatric Association, 14*(3), 270-277.

Webb, C., DuRubeis, R., & Barber, J. (2010). Therapist adherence/competence and treatment outcome: a meta-analytic review. *Journal of Consulting and Clinical Psychology, 78*, 200-211.

Webster-Stratton, C. (1998). Preventing conduct problems in Head Start children: Strengthening parenting competencies. *Journal of Consulting and Clinical Psychology*, *66*(5), 715–730. https://doi.org/10.1037/0022-006X.66.5.715.

Webster-Stratton, C., Reid, J., & Beauchaine, T. (2013). One-year follow-up of combined parent and child intervention for young children with ADHD.

Journal of Clinical Child & Adolescent Psychology, 42(2), 251-261. https://doi.org/10.1080/15374416.2012.723263

Wiersema, J., & Roeyers, H. (2009). ERP correlates of effortful control in children with varying levels of ADHD symptoms. *Journal of Abnormal Child Psychology, 37*, 327–336.

Yaruss, S. (2019). Direct therapy for early childhood stuttering: When, why and how? American Speech and Hearing Association, Orlando, FL.

Yaruss, S., Coleman, C., & Hammer, D. (2006). Treating preschool children who stutter: Description and preliminary evaluation of a family-focused treatment approach. *Language, Speech, and Hearing Services in Schools, 37*, 118-136. https://doi.org/0161-1461/06/3702-0118.

Yaruss, J. S., & Quesal, R. W. (2004). Stuttering and the International Classification of Functioning, Disability, and Health (ICF): An update. *Journal of Communication Disorders*, *37*, 35–52.

Zengin-Bolatkale, H., Conture, E., & Walden, T. (2015). Sympathetic arousal of young children who stutter during a stressful picture naming task. *Journal of Fluency Disorders, 46*, 24-40. https://doi.org/10.1016.j.jfludis.2015.07.005.

Zengin-Bolatkale, H., Conture, E., Key, A., Walden, T., & Jones, R. (2018). Cortical associates of emotional reactivity and regulation in childhood stuttering. *Journal of Fluency Disorders, 56*, 81–99.

Zengin-Bolatkale, H., Conture, E., Walden, T., & Jones, R. (2018b). Sympathetic arousal as a marker of chronicity in childhood stuttering. *Developmental Psychology, 43*(2), 135-151. https://doi.org/10.1080/87565641.2018.1432621.

Zhang, W. (2014). Mixed methods application in health intervention research: A multiple case study. *International Journal of Multiple Research Approaches, 8*(1), 24-35.

Appendices

Appendix A

Permission to Use Copyright Material

No permission statements required for inclusion of Papers 1 and 2.

Permission to include Paper 3:

Permissions Asha <Permissions@asha.org>
Wed 6/17/2020 1:04 AM
To: Kerianne Druker

Hi, Kerianne:

Thank you for providing the additional details. Permission is granted to reprint

JSLHR-19-00310R3
Title: An evaluation of an integrated stuttering and parent-administered self-regulation program for early developmental stuttering disorders

in your forthcoming dissertation. Please cite ASHA as the source and include a link to the original article online if it is available at the time of submission of your thesis. If your thesis will be published later, additional permission to reprint will be required at that time.

Best regards,

Libby

Appendix B

Author Attribution Statements

Attribution Statement for Paper 1- percentage contribution by field of activity:

	Conception and design	Data Collection and Method	Analysis and statistical method	Interpretation and discussion	Final Approval	Total % Contribution
Co-author 1 Neville Hennessey	✓		✓	✓	✓	10%
Co Author 1 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						
Co-author 2 Trevor Mazzucchelli	✓		✓	✓	✓	8%
Co Author 2 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						
Co Author 3 **Janet Beilby**	✓	✓		✓	✓	15%
Co Author 3 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						

Attribution Statement for Paper 2 - percentage contribution by field of activity:

|	Conception and design	Data Collection and Method	Analysis and statistical method	Interpretation and discussion	Final Approval	Total % Contribution
Co-author 1 Neville Hennessey	✔		✔	✔	✔	8%
Co Author 1 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						
Co-author 2 Trevor Mazzucchelli	✔	✔	✔	✔	✔	8%
Co Author 2 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						
Co Author 3 **Janet Beilby**	✔	✔		✔	✔	15%
Co Author 3 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						

Attribution Statement for Paper 3 - percentage contribution by field of activity:

	Conception and design	Data Collection and Method	Analysis and statistical method	Interpretation and discussion	Final Approval	Total % Contribution
Co-author 1 Neville Hennessey	✔		✔	✔	✔	8%
Co Author 1 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						
Co-author 2 Trevor Mazzucchelli	✔		✔	✔	✔	8%
Co Author 2 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						
Co Author 3 **Janet Beilby**	✔	✔		✔	✔	15%
Co Author 3 Acknowledgment I acknowledge that these represent my contribution to the above research output. Signed:						

Appendix C

Additional Publication during Candidature
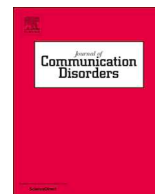
An evaluation of an integrated fluency and resilience program for early developmental stuttering disorders

Kerianne C. Druker[b], Trevor G. Mazzucchelli[a], Janet M. Beilby[b],[*]

[a] School of Psychology, Curtin University, Western Australia, Australia
[b] School of Occupational Therapy, Social Work and Speech Pathology, Curtin University, Western Australia, Australia

ABSTRACT

Purpose: The temperament construct of effortful control, an index of self-regulation and resilience, has been found to be predictive of stuttering severity in children and is a potential indicator of clinical prognosis. Evidence supports early intervention for preschool stuttering, and the successful effect of parents as agents of change in their children's stuttering therapy. However, no previous studies have trained parents to improve self-regulation in their children as a component of stuttering therapy. This study aimed to explore the effects of addressing self-regulation, as a component of stuttering treatment, on child fluency as well as parent and child psychosocial outcomes.
Methods: This study implemented a preliminary parent administered resilience component in conjunction with stuttering therapy for children who stutter, and compared outcomes to a cohort of children who stutter who received stuttering therapy only. Twenty-eight children who stutter were randomly allocated to one of the two treatment groups. Dependent variables tested pre- and post-treatment included stuttered speech severity, parenting practices, and child resilience indicators. Outcomes were compared between groups at post treatment.
Results: Stuttered speech severity decreased in both treatment groups. A reduction in behavioural and emotional problems, and increase in resilience was observed in the children who stutter whose parents received the additional resilience component of therapy. Furthermore, a significant improvement in parenting practices was demonstrated in this group. No significant changes in emotional and behavioural problems in children or parents were observed in the group of children who received fluency therapy only.
Conclusion: Results demonstrate that implementation of the resilience component was successful in positively shifting parenting practices and increasing behavioural resilience in children who stutter. This has clinical implications for successfully managing fluency while concurrently targeting the concomitant behavioural and emotional impacts of the disorder on both children and parents, potentially a key future prognostic indicator of the maintenance of fluency outcomes.

1. Introduction

Stuttering results in involuntary motoric interruptions to the forward flow of fluent speech production, impacting the successful delivery of communicative messages (Langevin, Packman, & Onslow, 2010). It is a multi-factorial disorder that encompasses a range of linguistic, motoric, environmental, psychosocial and personal factors that influence its onset, prognosis, severity and typography (Beilby, 2014; World Health Organization (WHO), 2001; Yaruss, 2010).

---

* Corresponding author.
  E-mail address: J.Beilby@curtin.edu.au (J.M. Beilby).

The International Classification of Functioning Disability and Health (ICF; World Health Organization (WHO), 2001) describes the comprehensive impact of the disorder on all facets of an individual's life experience, and recognises that the impact of stuttering extends far beyond an individual's exclusive speech features, manifesting in potentially long-term behavioural and social challenges, poor mental health outcomes, and impaired quality of life (Beilby, 2014; Conture, Kelly, & Walden, 2013; Vanryckeghem, Brutten, & Hernandez, 2005; Yaruss & Quesal, 2004). In addition, the ICF captures the impact of stuttering on family members (World Health Organization (WHO), 2001; Yaruss & Quesal, 2004). This is an important consideration as over 90% of parents of children who stutter (CWS) have reported emotional distress as a consequence of their children's stutter, including anxiety, sadness, guilt, self-blame, and uncertainty regarding their children's future (Langevin et al., 2010).

In light of these potentially broad psychosocial impacts on both CWS and their parents, delivering intervention as close to onset as possible is important to minimise the impact of the disorder, in terms of improved fluency and alleviation of child and parental concerns (De Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015; Franken, 2005). However, although therapeutic outcomes of early intervention stuttering therapies are encouraging, treatment is not universally successful for all children, with approximately one-quarter of preschool CWS only partially responding to treatment (De Sonneville-Koedoot et al., 2015). This variable success of early intervention cannot be predicated by gender, age, or socio-economic status (Kraft, Ambrose, & Chon, 2014; Manning & DiLollo, 2018). Conture et al. (2013) and Kraft et al. (2014) have suggested that another variable of interest is that of self-regulation.

The ecological model of resilience links resilience and self-regulation (Benzies & Mychasiuk, 2009). This model suggests that temperament characteristics are a key determinant of resilience capabilities (Benzies & Mychasiuk, 2009). Temperament refers to primarily genetically based differences in emotional reactivity and self-regulation (Rothbart, Ahadi, Hershey, & Fisher, 2001). Emotional reactivity refers to one's threshold for positive and negative arousal of emotions, and self-regulation refers to modulation of such reactivity (Eisenberg & Sulik, 2012). Consequently, resilient individuals control their emotional responses through exercising self-regulation (Hamill, 2003; Kesebir, Duru, Kucuksubasi, & Yaylaci, 2013). Optimal self-regulation occurs when problem solving abilities, coping strategies and emotional regulation are integrated to maximise one's capacity to respond to difficult situations (Hamill, 2003).

Studies have examined the temperament profiles of CWS and compared these profiles to those of children who do not stutter (CWNS). Such research provides evidence of the significant differences in temperament attributes between CWS and CWNS, specifically that CWS exhibit poorer abilities in regulation of emotions, and are consequently more emotionally reactive and less resilient (Conture et al., 2013; Johnson, Walden, Conture, & Karrass, 2010; Karrass et al., 2006; Schwenk, Conture, & Walden, 2007). Decreased self-regulation amongst CWS may manifest itself in greater difficulty exerting control over their complex speech motor systems to facilitate speech fluency (Kraft et al., 2014). In addition, this decreased emotional regulation and increased emotional reactivity presents behavioural and emotional problems (Eisenberg & Sulik, 2012).

The impact of temperament on stuttering severity was examined in Kraft et al. (2014). Of all factors tested in the study, including life events, home environment and temperament, the temperament construct of "effortful control", an index of self-regulation, on the Child Behaviour Questionnaire (CBQ; Rothbart et al., 2001) was the only significant factor found to be predictive of stuttering severity. Effortful control emerges in the first year of life and stabilises in early childhood, and comprises genetic and volitional components (Elizur, Somech, & Vinokur, 2016). Volitional components of effortful control are dependent on self-regulatory processes, and refer to the conscious inhibition of a dominant response for a desirable response (Eisenberg & Sulik, 2012; Kraft, Lowther, & Beilby, 2018). These self-regulatory processes link the ability to cope with personal distress and environmental changes, as children who are better emotionally regulated, and subsequently more resilient, are better at inhibiting or activating behaviours (Eisenberg & Spinrad, 2004).

Furthermore, a significant negative correlation was found between effortful control and stuttering severity, in that lower effortful control was predictive of a more severe stutter (Kraft et al., 2018). This indicates that CWS, particularly those with higher stuttered speech severity ratings, may have greater difficulty responding to environmental changes with natural self-regulation mechanisms, and are consequently challenged with increased stuttering behaviours (Kraft et al., 2014). Targeting these self-regulatory processes addresses the major components of multifactorial stuttering models, including the "Demands and Capacities Model" (Starkweather, 1987), as this model posits that CWS who are better able to deal with environmental stressors, will be less susceptible to breakdowns in speech fluency, potentially facilitating treatment success and maintenance. In addition, targeting these concomitant behavioural and emotional components of this disorder is efficacious in the context of the reported negative reactions to speech present in CWS as young as preschool age (Vanryckeghem et al., 2005).

Evidence indicating that children with more severe stutters are more emotionally reactive in response to changes in their external environment is important in the ecological model. This model posits that the availability of strong support networks in the external environment is crucial in fostering resilience (Benzies & Mychasiuk, 2009). Additional protective factors of interest, identified as positive determinants of resilience capabilities, include child emotional regulation and coping skills as well as supportive parent-child interactions (Benzies & Mychasiuk, 2009). This links closely with the stressor model described in Yaruss, Coleman, and Hammer (2006), that aims to identify specific factors affecting speech fluency. This model focuses on possible stressors within the child's overall development and temperament, some of which may be linked to effortful control, as well as possible stressors within the child's environment, including parent-child interactions and the predictability of the child's external environment. Therefore, resilience programs may target child self-regulatory processes in conjunction with support networks, such as parents or carers, in a child's daily environment (Benzies & Mychasiuk, 2009).

Parents of CWS have reported negative psychosocial consequences, uncertainty regarding stuttering management, as well as the "desire to do what is best for their children" (Plexico & Burrus, 2012, p. 275). Indirect approaches to stuttering therapy aim to empower parents to respond appropriately to their children's stuttering, and increase overall parental confidence in management

(Millard, Edwards, & Cook, 2009; Starkweather, 1987; Yaruss, 2010). As such, experts recognise that training parents in this regard provides skills to support their children's long-term speech fluency (Conture et al., 2013; Yaruss & Quesal, 2004).

In light of the positive impact of parent training in the management of early-onset stuttering, there may be significant treatment potential in training parents of CWS to improve resilience in their children while concurrently managing their stuttering (Elizur et al., 2016; Kraft et al., 2018; Pennington & Thomson, 2007). As such, changes to parenting practices may be a positive and vital element of stuttering therapy success and maintenance (Millard et al., 2009).

In summary, there is strong evidence of the significant differences in self-regulation, emotional reactivity, and resilience between CWS and CWNS. There is research supporting the effectiveness of early intervention for developmental stuttering. In addition, successful therapeutic outcomes have been demonstrated when parents are trained to facilitate fluency in their children. However, despite the research implicating enhanced self-regulation and resilience as a key factor of treatment success, there have been no previous studies examining the training of parents to improve self-regulation and behavioural resilience in CWS in early stuttering therapy.

The present study aimed to implement a parent-led resilience component in conjunction with early intervention stuttering therapy, and examine the impact of this integrated fluency and resilience program on fluency, parenting practices and child behavioural resilience outcomes. The resilience component incorporated aspects of training modules and activities from a variety of contemporary, published, efficacious resilience training programs for early childhood. A randomised control trial was implemented, with an enhanced treatment group that received the resilience component in conjunction with early intervention stuttering therapy (fluency plus resilience, FR), and an active unenhanced control group that received early intervention stuttering therapy only (fluency only, FO). Hypotheses are outlined below:

(a) Speech fluency will improve in both groups of children who stutter.
(b) CWS in the group receiving the additional resilience component will show an enhanced reduction to stuttered speech severity post treatment, when compared to the children receiving fluency therapy only.
(c) CWS in the group receiving the additional resilience component will show improvement in resilience indictors post treatment, including (a) reduction in behavioural and emotional difficulties (b) increased effortful control and (c) increased resilience characteristics.
(d) Parents receiving the resilience training will show improvements to parenting practices.
(e) No changes in resilience indicators or parenting practices will occur at post-treatment in the group of CWS who receive fluency therapy only.

2. Method

2.1. Research design

The study was a randomised control trial. Participants were randomly allocated to one of the two treatment conditions, being the enhanced treatment group receiving fluency therapy with an additional resilience component, and the comparison group receiving fluency therapy only with no additional resilience component. The study's methodology was approved by the Curtin University Human Research Ethics Committee.

2.2. Participants

Participants selected for this study were 31 dyads drawn from the Curtin University Stuttering Treatment Clinic (CSTC) waitlist comprising of a preschool CWS, aged from three to six years, and his/ her parent. A total of 25 participants were required to detect a large effect size ($\eta^2 = 0.25$), with 80% power, at a two-sided 5% significant level (Faul, Erdfelder, Buchner, & Lang, 2009).

CWS were eligible to participate if they met the following inclusion criteria: (a) diagnosis of developmental stuttering speech disorder by an experienced speech pathologist with over 10 years' experience in the management of stuttering disorders, (b) no history of speech, language, hearing and/or physiological impairments determined by parent report and investigator judgment in the initial assessment, (c) English as their primary language, no regular contact with another professional service for child behavioural problems. Parents were eligible to participate if they met the following inclusion criteria: (a) English as their primary language, (b) no intellectual impairment, (c) no regular contact with another professional service for parenting support. CWS were excluded based on the following criteria: (a) diagnosis of autism spectrum disorder, due to the unique temperament profiles of this disorder (Butean, Costescu, & Dobrean, 2014).

The majority of families (96%) were two parent co-parenting families living either together or separately but sharing custodial responsibilities. Four percent were single custodial parent families. All parents participated in the therapy and resilience intervention. The families were drawn from a range of lower, middle and higher socio-economic demographic areas with approximately 60% living in middle-income areas (Australian Bureau of Statistics, 2017).

Of 31 dyads recruited, three were not eligible to participate as the child presented with a language disorder, breach of inclusion criteria (ii), and were excluded from analyses (see Fig. 1). In total, 28 dyads met the criteria and were randomly allocated to group (14 to the FO group and 14 to the FR group). One dyad was lost to follow up in the FR group, resulting in 27 participants' data being analysed. No significant differences in age existed between groups (see Table 1 for mean ages in each group), confirmed by an independent samples t-test, $p > 0.5$. Participant demographics are included in Table 1.
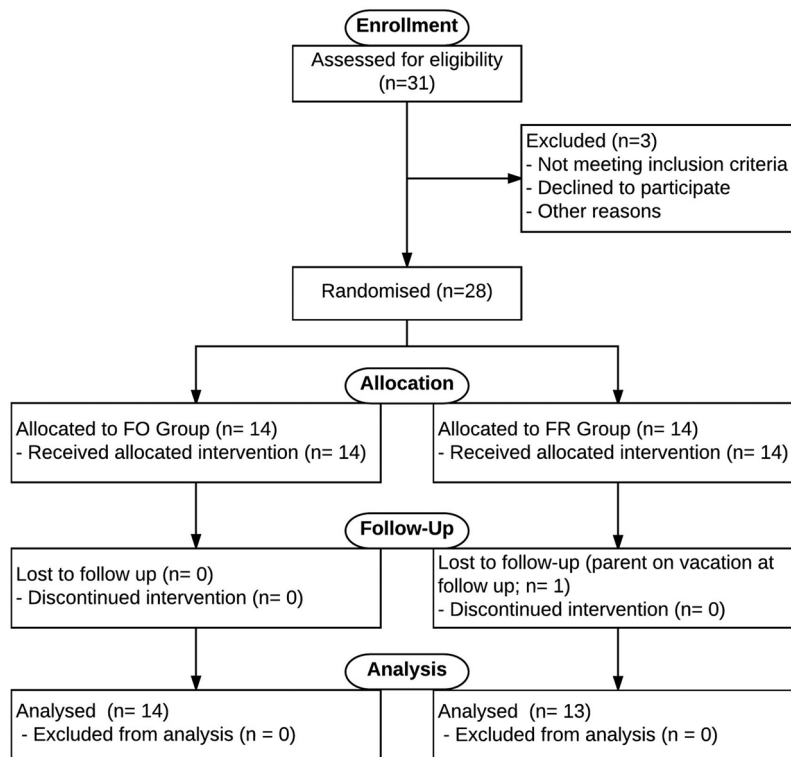
Fig. 1. CONSORT Flow Diagram.

Table 1
Participant Demographics.

	FO Group	FR Group
Average age in years (*SD*)	4.44 (0.99)	4.25 (1.02)
Number of males	9	7
Number of females	5	6
Mild stuttering severity pre-treatment (%SS)	2	4
Moderate stuttering severity pre-treatment (%SS)	5	3
Severe stuttering severity pre-treatment (%SS)	7	6

Note: severity is based on percentage syllables stuttered (%SS; Manning, 2010): mild 2–5, moderate 5–10, severe > 10.

2.3. Measures

Stuttered Severity Rating: Stuttering severity measures were taken pre- and post-therapy by raters blinded to the aims of the study. Stuttering severity ratings were collected by calculating the number of dysfluencies present per 100 syllables of speech using the Fluency Rater Application, i.e. the percentage of syllables stuttered (%SS) (ForFluency, 2012). Dysfluencies included whole-word and part-word repetitions, sound prolongations and blocks (Manning & DiLollo, 2018). Measures were collected during a conversational sample of 350–450 syllables in a time frame of approximately 30 min, while children were engaged in an informal play task (e.g. construction bricks or playdough) (Manning & DiLollo, 2018).

The Parenting and Family Adjustment Scales (PAFAS; Sanders, Morawska, Haslam, Filus, & Fletcher, 2014) is a comprehensive parent report inventory that has been used to evaluate parenting practices in a range of parenting intervention studies, including the Triple P Parenting Program (Sanders, 2012). The PAFAS consists of 30 items and measures five domains of parenting and family functioning that are known risk factors for behavioural and emotional difficulties in children (Shaw, Owens, Giovannelli, & Winslow, 2001). The PAFAS rating scales have strong internal consistency, with composite reliability estimates > .7 for all scales, and reported satisfactory predictive and construct validity (Sanders et al., 2014). At pre-treatment, there were no significant differences between groups in parenting practices, measured by the PAFAS, $t(1, 25) = 1.18$, $p = .25$.

The "parenting practices" domain was used to measure parenting outcomes in the study. Raw scores were added together from the following subscales: "parental consistency", "coercive parenting", "positive encouragement" and "parent-child relationship", providing an overall score for the "parenting practices" domain (Sanders et al., 2014). Items are scored on a 4-point Likert scale ranging

from "not true of me at all" (0) to "true of me very much, or most of the time" (3). Lower scores indicate more desirable parenting practices. The aim of the parent-administered resilience program was to encourage desirable parenting practices that reduce behavioural and emotional difficulties, and improve resilience in their children. Parents in both groups completed the PAFAS pre- and post-therapy, to measure the effect of the resilience program principles on enhancing desirable parenting practices.

The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) is a well-validated parent report measure examining behavioural and emotional problems, as well as strengths related to resilience. Children with increased emotional reactivity as a result of poor self-regulation abilities are known to have increased behavioural and emotional problems and less resilience characteristics, measured by the SDQ (Eisenberg & Sulik, 2012). An overall SDQ score was generated for each participant by summing scores from the following subscales: "emotional problems", "conduct problems", "hyperactivity" and "peer problems" (Goodman, 1997).

Two versions of the SDQ were administered depending on participant age (version one for children aged two to four years, and version two for children over four years). The mean Cronbach's alpha across subscales in both versions was > 0.7, indicating satisfactory internal consistency (Goodman, 1997). Across the two versions, 22 of the 25 items were identical. Wording of two items on the conduct scale, and one item on the hyperactivity scale, was softened for the younger age group. Parents in both groups completed the SDQ pre- and post-therapy, to measure the effect of the resilience program principles on behavioural and emotional stability, and strengths related to resilience in their children. Lower scores indicated fewer behavioural and emotional problems, and greater strengths related to resilience. At pre-treatment, there were no significant differences between groups in SDQ scores, $t(1, 25) = .97$, $p = 0.34$.

The effortful control subscale of the CBQ (CBQ-EC; Rothbart et al., 2001): The CBQ is a caregiver report measure examining temperament characteristics of preschool children aged three to seven years (Rothbart et al., 2001). It has been used to investigate the impact of temperament on stuttering severity, with the effortful control domain, an index of resilience, emerging as a significant predictor of stuttering severity (Kraft et al., 2018). In consultation with the authors of the CBQ, (M. Rothbart, personal communication, April 3, 2016) a version was created that included items measuring effortful control only, and comprised four subscales: "attention focusing", "inhibitory control", "low intensity pleasure", and "perceptual sensitivity". This abridged version consisted of 47 items, scored between 1 and 7 points, ranging from "extremely untrue of your child" (1) to "extremely true of your child" (7). Cronbach's alpha ranged from 0.64 to 0.78 for the scales, indicating satisfactory internal consistency (Rothbart et al., 2001). Parents in both groups completed this questionnaire pre- and post-therapy, to measure the effect of the resilience program principles on improving effortful control in their children. At pre-treatment, there were no significant differences between groups in CBQ-EC scores, $t(1, 25) = .20$, $p = 0.84$.

Curtin Early Childhood Stuttering Resilience Scale (CECSRS; Druker, Beilby, D'Orsogna, Thorogood, & Viljoen, personal communication, May 10, 2016): Specific measures of resilience in the preschool population are limited, therefore this exploratory scale was developed using the Delphi method from a focus group comprising the researcher and four experienced clinicians working in the field of stuttering disorders (Okoli & Pawlowski, 2004). This pilot measure, in the form of a parent checklist, constituted a set of observations regarding childhood behaviours that were specifically targeted in the parent-led resilience program. Items were drawn from the literature focusing on general resilience in the preschool population (Dent, 2016; Masten, 2001; Ungar, 2015). The content and structure of the CECSRS was based on the Grit scale (Duckworth & Quinn, 2009), a well-validated scale that measures resilience in the adolescent population. The CECSRS contained 9 items (see Appendix A for the scale and scoring instructions). Items were rated on a 5-point Likert scale ranging from "not at all like my child" (1), to "very much like my child" (5).

Initial validity testing was performed, by testing the relationship between CECSRS scores and scores on well-validated measures of similar constructs, including the SDQ and CBQ-EC. A negative, moderate, non-significant correlation was found between CECSRS and the SDQ, $r(27) = -0.31$, $p = .11$. Although this correlation was non-significant, the relationship was in the predicted direction, as children with greater resilience were expected to have fewer behavioural and emotional problems. A positive, moderate, non-significant correlation was found between CECSRS and CBQ-EC, $r(27) = .32$, $p = .10$. Again, although non-significant, the relationship was in the predicted direction as effortful control is considered an index of resilience. Parents in both groups completed this scale pre- and post-therapy, to measure the effect of the resilience program on promoting behavioural resilience characteristics in their children. At pre-treatment, significant differences were found to exist between groups in CECSRS scores, $t(1, 25) = 2.56$, $p = 0.02$, with CWS in the FR group having significantly higher scores (see Table 2). Therefore, changes observed on this scale from pre to post

Table 2
Simple Effects Analyses.

	FR			FO		
	Mean (*SD*)			Mean (*SD*)		
	Pre	Post	Sig	Pre	Post	Sig
PAFAS	8.54 (3.05)	5.15 (3.31)	0.01*	6.71 (4.87)	6.86 (3.41)	0.88
SDQ	12.46 (5.48)	8.31 (6.16)	< 0.01**	10.36 (5.72)	9.07 (4.86)	0.34
CECSRS	27.54 (4.16)	33.77 (3.81)	< 0.01**	32.07 (5.03)	32.64 (3.18)	0.63

Note: PAFAS = parenting and family adjustment scale, SDQ = Strengths and Difficulties Questionnaire, CECSRS = Curtin Early Childhood Stuttering Resilience Scale, *p < 0.05, **p < 0.01.

treatment were interpreted with caution, given a significant between group difference was already present before the intervention commenced.

2.4. Intervention

Beilby Stuttering Therapy (BST): BST administered at the CSTC is based on a combination of direct and indirect stuttering therapy. Direct therapy, in the form of response contingent principles, includes: drawing attention to the stutter using neutral terminology such as "bumps" and redirecting the child to produce fluent speech, such as "just once". Response contingencies are integrated with indirect treatment approaches including components of parent-child interaction therapy (Millard et al., 2009), whereby parents are trained to facilitate fluency in their children; and "Demands and Capacities Model" treatment (Starkweather, 1987), whereby motoric, cognitive and linguistic domains are manipulated to increase the child's capacity for fluency. Parents in both groups were trained in these techniques and implemented them throughout their daily conversation interactions with their children.

The Curtin Early Childhood Stuttering Resilience Program (Druker, Beilby, D'Orsogna, Thorogood, & Viljoen, personal communication, May 10, 2016): Constructs targeted in the resilience program were broad-based and drawn from needs identified in early stuttering clinical practice, techniques identified in the "Ordinary Magic" resilience research (Masten, 2001), as well as contemporary resilience research presented in seminars and presentations by Michael Ungar (Ungar, 2015) and Maggie Dent (Dent, 2016). The researcher and four speech pathologists with over 10 years' experience in the management of stuttering were involved in focused topic discussions, from which consensus was reached regarding areas that should be addressed to improve behavioural management and volatility in the children, with the aim of concurrently facilitating online fluency benefits (Okoli & Pawlowski, 2004). Broad areas targeted included: child independence, problem solving, responsibilities, problem ownership, setting firm limits and boundaries, social graces, parent encouragement versus praise, as well as parent self-nurturing; with the overarching aim of turning the frame of reference for acceptable behaviour back to the children, to improve their ability to manage and control their behavioural responses in the environment (see Appendix B for activity examples).

A set of integrated and applied resilience principles were presented verbally by the researcher, and in a written handout summary to parents in the FR group, in a 30-minute training session. Parents were asked to apply the resilience principles from week 3 to week 12 of therapy, which were to be embedded into daily activities in the home. Parents were given a logbook to track implementation of the resilience program principles and activities on a weekly basis (see Appendix C for an example page of the logbook). The supervising clinician at the CSTC provided additional weekly support to parents in implementation (see Appendix D). This clinician was involved in the design of the program and was present at every resilience training session. Behavioural fluency therapy was implemented and guided concurrently as part of the integrated fluency and resilience treatment program.

Delivery of therapy and techniques was encouraged to be conducted in a natural manner with frequency depending on the opportunities and/or challenges posed by the child throughout the day. Parents reported that following the initial training and with ongoing provision of individual support by the clinicians, their administration of the strategies and techniques became a regular part of their daily routine.

2.5. Procedure

Thirty-one participants were drawn from the CSTC waitlist. All CWS received a weekly 1-hour behavioural stuttering therapy session over a 12-week treatment block. In week 1 of therapy attendance, parents were provided a package including the following parent-report questionnaires which took a maximum of 30 min to complete: PAFAS (Sanders et al., 2014), SDQ (Goodman, 1997), CBQ-EC (Rothbart et al., 2001), and CECSRS. Parents were requested to complete and bring the questionnaires to the treatment session in week 2. Before each child's session in week 2, participants were randomly allocated to the FO or FR group.

If the child was allocated to the FR group, a 30-minute resilience training session was scheduled with his/her parent between weeks 2 and 3 of the treatment block. In these sessions, the principles of the resilience program were presented and parents were given a logbook to track the implementation of these principles. Over the 12-week treatment block, the supervising clinician at the clinic followed up with parents regarding implementation of the resilience principles and activities on a weekly basis, for an additional 5–10 minutes following their hourly fluency session. This information was documented in each child's progress notes for future evaluation. In week 11, the original package of questionnaires was re-issued to all parents to be returned in week 12.

Of the 27 participant dyads who completed treatment, 23 received the target 12 intervention sessions within the allocated concurrent 12-week treatment block. Three participants received the 12 sessions over 13 concurrent weeks (with one postponed session to the following week) and one participant over 14 concurrent weeks (with two postponed sessions to following weeks), as a result of rescheduling due to child illness.

3. Results

3.1. Data analysis

Five two-way mixed ANOVAs with a two-tailed alpha level of 0.05 were conducted to measure pre-post treatment change on each dependent variable in both groups, as well as to test for an interaction between time and group. No significant outliers were identified, after reviewing the data.
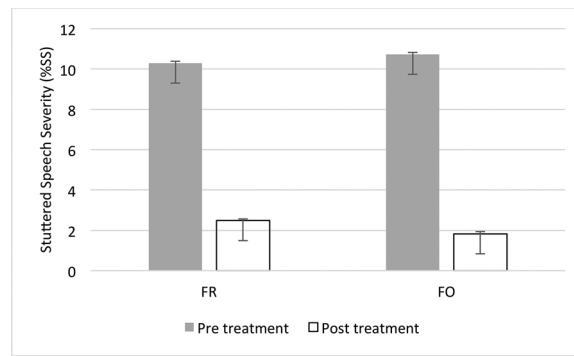
Fig. 2. Mean stuttering severity scores (%SS) at pre-treatment and post-treatment in the fluency only (FO) and fluency plus resilience (FR) group. *Note:* Error bars represent the standard error of the mean.

All data were normally distributed except for: post-treatment %SS scores in the FR group, $W(13) = 0.61, p < .01$, pre-treatment PAFAS scores in the FO group, $W(14) = 0.82, p = .01$, and post-treatment SDQ scores in both the FO group, $W(14) = 0.86, p = .03$, and the FR group $W(13) = 0.86, p = .04$. For these data sets with significant Shapiro-Wilk statistics (excluding post-treatment %SS scores in the FR group), z scores for skewness were not excessive $-3 < z < 3$, therefore data was acceptable for ANOVA (Allen & Bennett, 2012). The z score for skewness was > 3 for %SS scores in the FR group, therefore %SS scores were transformed using log^10 ratio as distributions were positively skewed (Allen & Bennett, 2012). Following this transformation, $-3 < z < 3$ for all %SS data sets, acceptable for ANOVA (Allen & Bennett, 2012).

Homogeneity of variance assumptions were evaluated using Levene's test statistics. This assumption was met for all data sets, excluding pre-treatment PAFAS scores. ANOVA is robust to violation of equal variances, as the sample is moderately sized and groups were approximately equal (Allen & Bennett, 2012), therefore this data set was acceptable for ANOVA. Sphericity testing was irrelevant as the repeated measures factor had only two levels for all ANOVAs conducted (Allen & Bennett, 2012). Descriptive statistics were computed for each data set.

For stuttering severity (%SS), the main effect of time was statistically significant, $F(1, 25) = 100.99, p < .01$, large effect size, $\eta^2 = .80$, with stuttering severity (%SS) significantly lower at post-treatment ($M = 2.14, SD = 0.32$) than at pre-treatment ($M = 10.47, SD = 0.32$), consistent with the corresponding hypothesis. The main effect of group was non-significant, $F(1, 25) = .31$, $p = .58$. The interaction between group and time was non-significant, $F(1, 25) = 1.23, p = .28$, inconsistent with the corresponding prediction that children in the FR group would show an enhanced reduction to stuttered speech severity post treatment, compared to the children who received fluency therapy only (Fig. 2).

For parenting practices (PAFAS), the main effect of time was significant, $F(1, 25) = 4.78, p = .04$, large effect size, $\eta^2 = .16$, with scores at post-treatment ($M = 6.01, SD = 3.41$) significantly lower than at pre-treatment ($M = 7.63, SD = 4.13$). Note lower scores indicate more desirable parenting practices. The main effect of group was non-significant, $F(1, 25) < 0.01, p = .96$. The interaction between group and time was significant, $F(1, 25) = 5.66, p = 0.03$, large effect size, $\eta^2 = .18$ (see Fig. 3). Simple effects analyses were conducted to examine this significant interaction, and revealed that the effect of time on parenting practices was significant in the FR group, $F(1, 12) = 8.8, p = .01$, large effect size, $\eta^2 = .42$, with scores higher at pre-treatment than post-treatment, consistent with the corresponding hypothesis (see Table 2 for difference in means). This effect was non-significant in the FO group, $F(1, 13) = .02, p = .88$, with no trend evident.

For behavioural and emotional problems and strengths related to resilience i.e. strengths and difficulties (SDQ), the main effect of time was significant, $F(1, 25) = 12.7, p < .01$, large effect size, $\eta^2 = 0.34$, with SDQ scores at post-treatment ($M = 8.69, SD = 5.43$)
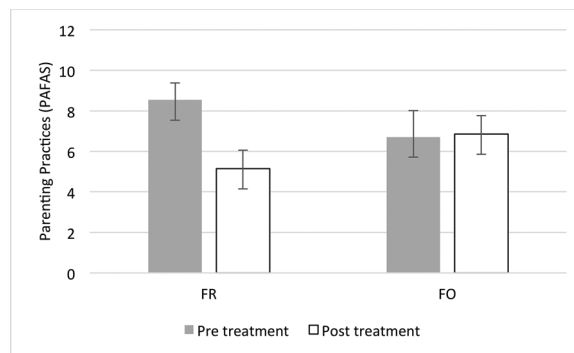
Fig. 3. Mean "parenting practices" scores at pre-treatment and post-treatment in the fluency only (FO) and the fluency plus resilience (FR) group. *Note:* Error bars represent the standard error of the mean.

Fig. 4. Mean "strengths and difficulties" scores at pre-treatment and post-treatment in the fluency only (FO) and the fluency plus resilience (FR) group. *Note:* Error bars represent the standard error of the mean.
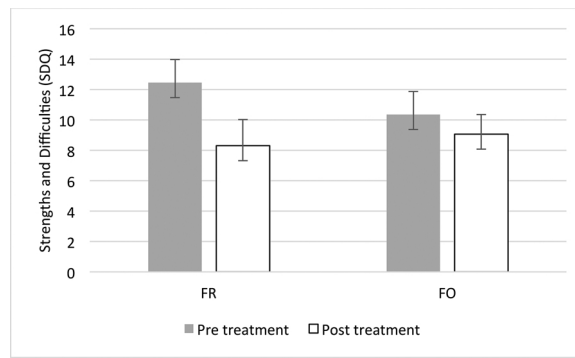
significantly lower than at pre-treatment ($M = 11.41$, $SD = 5.60$). Note lower scores indicate fewer behavioural and emotional problems, and greater strengths related to resilience. The main effect of group was non-significant, $F(1, 25) = .11$, $p = .74$. The interaction between group and time was approaching significance, $F(1, 25) = 3.53$, $p = .07$ (see Fig. 4). As this interaction was in line with a directional hypothesis, simple effects analyses were conducted, indicating that the effect of time on SDQ scores was significant in the FR group, $F(1, 12) = 30.7$, $p < .01$, large effect size, $\eta^2 = .72$, with SDQ scores higher at pre-treatment than post-treatment, consistent with the corresponding hypothesis (see Table 2 for difference in means). Although a similar decreasing trend occurred in the FO group, this effect was non-significant $F(1, 13) = .99$, $p = 0.34$.

For effortful control (CBQ-EC), the main effects of time, $F(1, 25) = .20$, $p = .66$, and group, $F(1, 25) = .09$, $p = .77$, and the interaction between time and group, $F(1, 25) = .81$, $p = .38$, were not significant (see Fig. 5). These findings do not support the corresponding hypothesis for the FR group.

For resilience characteristics (CECSRS), the main effect of time was significant, $F(1, 25) = 15.36$, $p = < 0.01$, large effect size, $\eta^2 = .38$, with a significant increase in resilience scores from pre-treatment ($M = 29.8$, $SD = 5.09$) to post-treatment ($M = 33.21$, $SD = 3.48$). The main effect of group was non-significant, $F(1, 25) = 1.66$, $p = .21$. There was a significant interaction between group and time, $F(1, 25) = 10.63$, $p < .01$, large effect size, $\eta^2 = .30$ (see Fig. 6). Follow up analyses revealed that the effect of time on resilience was significant in the FR group, $F(1, 12) = 23.27$, $p < 0.01$, large effect size, $\eta^2 = .66$, i.e. resilience scores were significantly higher post-treatment than pre- treatment, consistent with the corresponding hypothesis (see Table 2 for difference in means). No trend was found in the FO group, $F(1, 13) = .24$, $p = 0.63$.

Bivariate Pearson's correlations were run to assess the size and direction of any relationship between stuttering severity and other dependent variables post-treatment. The z scores for skewness were not excessive $-3 < z < 3$ after transformation of %SS scores, therefore the assumption of normality was met (Allen & Bennett, 2012). The assumptions of linearity and homoscedasticity were supported. The bivariate correlation between stuttering severity and effortful control was negative, medium, and significant, $r(25) = -0.38$, $p = .05$. The bivariate correlations between stuttering severity (%SS scores) on one hand, and parenting practices (PAFAS scores), behavioural and emotional stability (SDQ scores), and resilience characteristics (CECSRS scores) on the other were all non-significant, $ps > .05$.

4. Discussion

According to the Speech Pathology Australia Code of Ethics (Speech Pathology Australia, 2010), speech pathologists are obligated to work to improve the quality of life in the child who stutters, by addressing the speech impairment, as well as concomitant
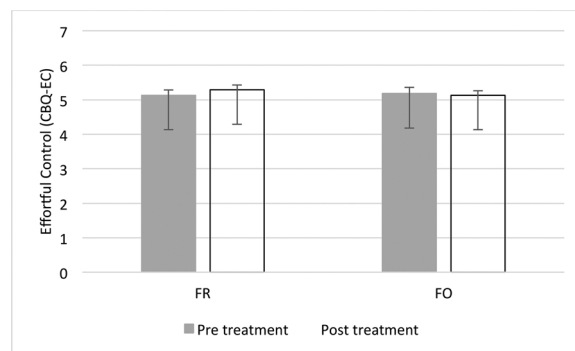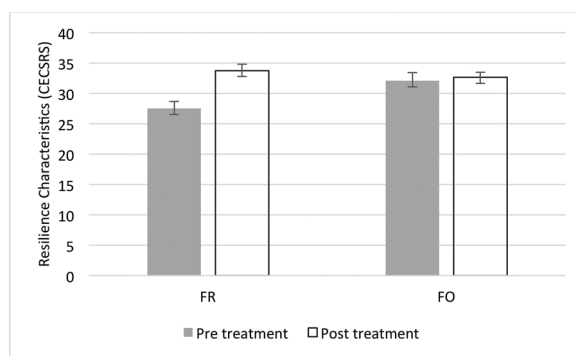

Fig. 5. Mean effortful control scores at pre-treatment and post-treatment in the fluency only (FO) and fluency plus resilience (FR) group. *Note:* Error bars represent the standard error of the mean.

Fig. 6. Mean "resilience characteristics" scores at pre-treatment and at post-treatment in the fluency only (FO) and the fluency plus resilience (FR) group. *Note:* Error bars represent the standard error of the mean.

emotional and behavioural factors that may impact a child's activities and participation (World Health Organization (WHO), 2001). Addressing these emotional and behavioural factors to facilitate prognostic gains was a focus of this study. The study's research hypotheses are addressed below.

4.1. Speech fluency will improve in both groups of children who stutter

The demonstrated significant pre-post reduction in stuttered speech severity highlights the importance of delivering early intervention as close to onset as possible, to enhance fluency and subsequently alleviate the possible negative psychosocial impact for CWS and their parents (Yaruss & Quesal, 2004).

4.2. CWS in the group receiving the additional resilience component will show an enhanced reduction to stuttered speech severity post treatment, when compared to the children receiving fluency therapy only

Given that this hypothesis was not supported, the authors consider that the time span of the treatment block may have been too short to capture such changes. In a randomised control trial examining the effectiveness of early stuttering intervention, 25–30% of preschool CWS did not maintain gains in improved fluency at 18 months follow up (De Sonneville-Koedoot et al., 2015). Therefore, targeting these behavioural components in CWS may be a possible significant prognostic indicator of maintenance of fluency gains; potentially yielding enhanced effects on fluency in the FR group at follow up, if monitored longitudinally (Kraft et al., 2014).

In addition, if all children presented with severe stutters pre-treatment, CWS in the FR group may have been more receptive to the resilience program principles, and shown immediate enhanced improvement to fluency as a result. This is supported by the significant negative correlation found between stuttering severity and effortful control post-treatment, showing children with more severe stutters to have lower effortful control. This finding endorses contemporary stuttering research, whereby the same correlation between stuttering severity and effortful control was found (Kraft et al., 2018). More immediate effects of addressing self-regulation on speech fluency may be captured by providing self-regulation training to CWS who specifically present with self-regulation challenges. The fluency capacities of this subgroup of CWS may be more quickly impacted by improved self-regulation, as relatively greater emotional and temperament demands are lessened (Starkweather, 1987).

4.3. CWS in the group receiving the additional resilience component will show improvement in resilience indictors post treatment, including (a) reduction in behavioural and emotional difficulties (b) increased effortful control and (c) increased resilience characteristics

(a) Negative emotions associated with stuttering, present at preschool age, may evolve into emotional and behavioural problems, and if not addressed could lead to significant mental health challenges (Vanryckeghem et al., 2005; Yaruss & Quesal, 2004). The demonstrated significant reduction in SDQ scores over the short 12-week treatment block is highly significant for this at-risk population, providing direction to improve mental health outcomes for these children from a very young age; once again supporting the importance of early intervention in stuttering management.

(b) This component of the hypothesis was not supported. Previous research revealed that effortful control is a predictor of stuttering severity (Kraft et al., 2014), however no previous research has attempted to monitor change in effortful control over the course of therapy. Effortful control is primarily a genetically based temperament characteristic, which may be difficult to shift (Kraft et al., 2018). Therefore, major shifts in effortful control would have to occur to demonstrate statistically significant changes on a parent-report measure, unlikely to be achieved in a short duration of the 12-week treatment block. As effortful control is dependent on self-regulatory processes, the significant improvement in self-regulation skills shown on the SDQ, and increase in resilience characteristics on the CECSRS, may eventuate into statistically significant change in the overall temperament construct of

effortful control, if monitored longitudinally. In addition, the trajectory of positive shifts in parenting practices may support significant positive changes in effortful control in these children in the future (Elizur et al., 2016; Sanders et al., 2014).

(c) Although this was a pilot scale, the scale captured a significant improvement in resilience characteristics in the FR group. These results were consistent with those yielded by the SDQ, a standardised measure of similar constructs. Furthermore, in this study's intervention designed to specifically target behavioural resilience in the preschool population, significant improvement in resilience characteristics was demonstrated on this scale, supporting the scale's initial content validity. However, further reliability and validity testing of the scale is required to support the validity of results yielded on this scale. In addition, the between group differences in CECSRS scores present at pre-treatment, indicate that the lack of change in CECSRS scores in the FO group may be at least in part due to their scores being higher at pre-treatment.

4.4. Parents receiving the resilience training will show improvements to parenting practices

The demonstrated significant positive shift in overall parenting practices in the FR group is underscored by the ecological model of resilience, which highlights the importance of external supports in training of childhood resilience (Benzies & Mychasiuk, 2009).

4.5. No changes in resilience indicators or parenting practices will occur at post-treatment in the group of CWS who receive fluency therapy only

Outside of improved stuttered speech severity, there were no other significant changes in the FO group. These non-significant findings underscore the internal validity of study findings, indicating that significant changes on these outcomes in the FR group can be attributed to the exclusivity and implementation of the resilience program principles.

4.6. Future implications

This study provides initial data supporting the concept that the self-regulation component of resilience can be improved. Self-regulation as an adjunct to therapy has recently emerged as a significant prognostic indicator for stuttering (Kraft et al., 2014). Poor self-regulation consumes resources in terms of parenting time, emotional volatility on the part of the child, and disrupted structure in the family throughout the day (Goodman, Ford, Simmons, Gatward, & Meltzer, 2000). In contemporary therapy, time is a challenge and parents require support (Langevin et al., 2010). Therefore, the implementation of the resilience program principles in conjunction with early intervention stuttering therapy has positive implications for these practice considerations.

In addition, improvement in self-regulation and resilience addresses the major components of multifactorial stuttering models including the "Demands and Capacities Model" (Starkweather, 1987) that posits children who are more resilient and subsequently better equipped to deal with environmental stressors with less emotional volatility, will have increased resources to devote to facilitating fluent speech production. However, given there were no differences in speech fluency between groups at post-treatment, clinical implications in the context of immediate improvements to speech fluency may be premature. However, the resilience program principles implemented in this study may have future implications for stuttering therapy, in terms of improving the behavioural component of temperament to enhance the maintenance of treatment outcomes.

A further hallmark finding of this study was a positive shift in parenting practices. Training parents with the resilience program principles addressed the need for increased parent support, as the shift in positive parenting likely resulted in parents who felt more confident and empowered in their overall parenting practices (Sanders et al., 2014). A cornerstone of evidence-based practice considers client factors in treatment implementation (Gillam & Gillam, 2008). Therefore, considering the family as the definition of client, and providing parents with positive direction and support in the management of this complex disorder, is efficacious in the context of reported parent distress and anxiety regarding their children's stuttering management (Langevin et al., 2010). In addition, these positive parenting practices will likely map onto a positive trajectory of wellbeing for these children and their families, with improved child and parent mental health outcomes reported as a result of desirable parenting practices over time (Kazdin & Blase, 2011).

According to the Speech Pathology Australia Code of Ethics, clinicians are charged with managing the wellbeing of the child and family, beyond targeting the communication disorder in isolation (Speech Pathology Australia, 2010). This study was an initial investigation that showed improvement in the potential concomitant behavioural and emotional impacts of the developmental stuttering disorder in both CWS and their parents, beyond isolated improvements in stuttered speech severity (Vanryckeghem et al., 2005). As such, the findings address developmental stuttering from a holistic perspective, providing support for the implementation of resilience principles as a potentially vital additional component of early intervention stuttering therapy (World Health Organization (WHO), 2001).

4.7. Limitations

Evidence for the reliability of parent report measures has shown them to be a valid assessment of temperament (Rothbart et al., 2001). However, potential informant bias may have been present, as parent report questionnaires were used in isolation as measures of observed parenting practices and child behaviour. Additional clinical observations would further augment the validity of study

findings.

The 12-week treatment block may have been too short to capture all potentially significant results. Longitudinal follow up is recommended to examine if changes to parenting practices and improved self-regulation in the children in the FR group eventuate into changes to effortful control, and to examine the impact of the resilience component in the maintenance of fluency and behavioural resilience outcomes over time.

Further reliability and validity testing is required to support the validity of the findings yielded on the pilot CECSRS. Although preliminary validity testing was carried out, no significant correlations were found between scores on this scale and stuttered speech severity. This poses a possible challenge to the validity of this scale as a measure of self-regulation, as children with poorer self-regulation skills characterised by low effortful control have been found to have increased stuttered speech severity (Kraft et al., 2014, 2018).

The resilience training implemented in the study was not standardised. Implementing an evidence-based parenting intervention that has been found to improve self-regulation skills in children would increase the replicability of the study. Furthermore, given that not all children who stutter exhibit low self-regulation skills (Donaher & Richels, 2012; Druker, Hennessey, Mazzucchelli, & Beilby, 2019), future integrated interventions addressing self-regulation as a component of stuttering treatment may be more suited to CWS specifically identified to present with concomitant low self-regulation skills.

4.8. Directions for future research

The nature of the clinical trial conducted in the study was exploratory. Strengths of the study were randomisation to group, minimal loss to follow up, and the use of valid quantitative measures to demonstrate change (SDQ and PAFAS). This study can be viewed as a preliminary foundation for replication and extension with a larger randomised clinical trial, with increased sample size and study power to detect statistically and clinically significant results. Wider recruitment of participants and clinicians from different clinical settings is also recommended to examine diverse early intervention programs beyond the CSTC. In addition, recruitment of children with more severe stuttered speech behaviours is recommended to increase the opportunity to examine correlation effects further.

Future qualitative research is recommended to gain insight into parents' experience implementing the resilience principles, and to achieve a more in-depth understanding of the impact of these principles on their parenting, as well as their children's behaviour and fluency in daily life through their narrative reflections (Kazdin & Blase, 2011). In addition, clinical changes in measures that are valid predictors of child and family wellbeing have been revealed in this study (Goodman et al., 2000; Sanders et al., 2014). Therefore, longitudinal follow up is recommended to measure maintenance and durability of these positive changes, as well as to detect any further longitudinal trajectory of change over time.

5. Conclusion

This study provides evidence for the overall effectiveness of early intervention in stuttering therapy. Beyond the fluency aspect of stuttering intervention, the resilience program principles were successful in positively shifting parenting practices, and developing improved self-regulation and resilience in CWS. This positive shift in parent practices and increased child behavioural resilience will enable children to deal with environmental changes with less behavioural and emotional volatility, likely supporting long-term maintenance of fluency outcomes (Kraft et al., 2014). Furthermore, this may result in a trajectory of improved mental health outcomes for both children and parents in this vulnerable population (Beilby, 2014; Goodman et al., 2000; Langevin et al., 2010).

CRediT authorship contribution statement

Kerianne C. Druker: Conceptualization, Methodology, Formal analysis, Investigation, Writing - original draft, Visualization. **Trevor G. Mazzucchelli:** Conceptualization, Methodology, Writing - review & editing, Supervision. **Janet M. Beilby:** Conceptualization, Methodology, Writing - review & editing, Supervision.

Appendix A

Curtin Early Stuttering Childhood Resilience Scale

Please respond to the following 9 items. Examples are provided where necessary.

1. My child is aware that there are consequences for his/her actions.
e.g. if he/she breaks something, does he/she try fix it?

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

2. My child usually finishes an activity that he/she starts.
e.g. if your child begins a jigsaw puzzle, is he/she likely to finish it?

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

3. My child frequently reacts negatively by getting upset and angry, when he/she hears "no" or doesn't get his/her way.
e.g. your child gets upset when you don't agree to buy him/her a treat at the shops.

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

4. My child is responsible for doing simple chores around the house.
e.g. helping to set the table or pack away his/her toys

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

5. My child is able to solve problems.
e.g. if a favourite toy gets stuck underneath a table, your child will show initiative to retrieve it.

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

6. My child is clingy.

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

7. My child enjoys sharing toys with other children.

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

8. When my child falls over or hurts him/herself, he/she quickly gets over it.

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

9. My child is relatively independent.
e.g. takes some responsibility for dressing him/herself in the morning before school.

☐Very much like my child
☐Mostly like my child
☐Somewhat like my child
☐Not much like my child
☐Not at all like my child

Scoring Key - Curtin Early Stuttering Childhood Resilience Scale

Scoring for items 1, 2, 3, 5, 7, 8, 9:
- Very much like my child – 5 points
- Mostly like my child – 4 points
- Somewhat like my child – 3 points
- Not much like my child – 2 points
- Not at all like my child at all – 1 point

Scoring for items 4, 6:

- Very much like my child – 1 point
- Mostly like my child – 2 points
- Somewhat like my child – 3 points
- Not much like my child – 4 points
- Not at all like my child at all – 5 points

Higher scores indicate greater resilience

Appendix B

Example Activity for Each Area Targeted in the Resilience Program.

Area		Example Activity
1	Independence	Child to dress him/herself every morning
2	Problem Solving	If a favourite toy gets stuck underneath a table, encourage the child to brainstorm how to retrieve it
3	Responsibility	Set daily chores e.g. make own bed and help set the dinner table
4	Problem Ownership	Acknowledge behaviour is problematic and apologise to parent/sibling/friend e.g. shouting at parent
5	Firm limits and boundaries	Consistent parent response to child's behaviour, time out for unacceptable behaviours such as physical violence
6	Social Graces	Child is encouraged to wait until parent is finished talking or say "excuse me" (online fluency benefit)
7	Parent encouragement	If a child is making a puzzle, provide encouragement: "you seem so proud of what you have made" vs. praise: "you're so clever"
8	Parent self-nurturing	Parent given "permission" to take 30 minutes of time out during the day e.g. listen to music while children play in another room

Appendix C

Example Page in the Logbook Provided to Parents to Track Implementation of the Resilience Program Principles
WEEK 1

	Activity	Tally	Comments/questions
Independence	*1. Getting dressed*	IIII	*Even helped his brother with his socks after he dressed himself!*
	2. Taking lunch box out of school bag and putting on bench	IIII	*He enjoyed this responsibility (needed 3 reminders)*
	3. Putting clothes in laundry basket	IIII	*He enjoyed this responsibility (needed 3 reminders)*
Problem Solving	*1. Rearranging Lego to fit all the blocks into the container in order to close the lid*	I	*I would usually have done this without involving him.*
	2. Fitting a puzzle piece into the correct place	IIII	*I would usually place it in the spot for him without encouraging him to problem solve*
	3. Child asks for help when shoelaces come undone	III	*Usually continues playing/ running around when shoelaces come undone*
Problem Ownership	*1. Saying sorry for hitting brother before being told to*	IIII	*Usually requires lots of reinforcement*
	2. Asking for help instead of whining when he has a problem	III	*I usually give in to the whining and fix the problem for him without encouraging him to use his words*
	3. Apologising for not packing away toys after brother tripped over them	I	*Impressed he was able to do this spontaneously*

Appendix D

Questions Guiding Weekly Support Provided to Parents in the Fluency plus Resilience (FR) Group

1 How are you feeling about putting these resilience principles into place on a daily basis?
2 What particular resilience principle/s have you been focusing on with your child in the last week? Give examples of situations/ times when you have implemented these.
3 Are they having any impact on your parenting? If yes, please describe this impact.
4 How did your child respond?
5 Have you noticed any change in your child's behaviour over the last week? Please give examples.
6 If yes, has this change in behaviour affected his/ her fluency (in a positive or negative way)? In what situations?
7 What are you going to continue to focus on this week? What will you change?
8 Do you have any other questions about the resilience program that you have recorded in your logbook?

References

Allen, P., & Bennett, K. (2012). *SPSS statistics: A practical guide version 20* (1st ed.). South Melbourne, VIC: Cengage Learning Australia.

Australian Bureau of Statistics (2017). *Household income and wealth, Australia, 2015–16. [Survey].* Retrieved from http://www.abs.gov.au.

Beilby, J. (2014). Psychosocial impact of living with a stuttering disorder: Knowing is not enough. *Seminars in Speech and Language, 35*, 132–143. https://doi.org/10.1055/s-0034-1371756.

Benzies, K., & Mychasiuk, R. (2009). Fostering family resiliency: A review of the key protective factors. *Child and Family Social Work, 14*(1), 103–114. https://doi.org/10.1111/j.1365-2206.2008.00586.x.

Butean, I., Costescu, C., & Dobrean, A. (2014). Differences between empathic responses in children with autism spectrum disorder and typically developing children. *Journal of Evidence-Based Psychotherapies, 14*(2), 197–209. Retrieved from http://catalogue.curtin.edu.au/.

Conture, E. G., Kelly, E. M., & Walden, T. A. (2013). Temperament, speech and language: An overview. *Journal of Communication Disorders, 46*(2), 125–142. https://doi.org/10.1016/j.jcomdis.2012.11.002.

De Sonneville-Koedoot, C., Stolk, E., Rietveld, T., & Franken, M. (2015). Direct versus indirect treatment for preschool children who stutter: The RESTART randomized trial. *PloS One, 10*(7), 1–17. https://doi.org/10.1371/journal.pone.0133758.

Dent, M. (2016). *Maggie Dent: Quietly improving lives.* Retrieved from http://www.maggiedent.com/content/seminars-parents.

Donaher, J., & Richels, C. (2012). Traits of Attention Deficit/Hyperactivity Disorder in school-age children who stutter. *Journal of Fluency Disorders, 37*(4), 242–252.

Druker, K. C., Hennessey, N., Mazzucchelli, T. G., & Beilby, J. M. (2019). Elevated attention deficit hyperactivity disorder symptoms in children who stutter. *Journal of Fluency Disorders, 59*, 80–90. https://doi.org/10.1016/j.jfludis.2018.11.002.

Duckworth, A. L., & Quinn, P. D. (2009). Development and validation of the Short Grit Scale (Grit- S). *Journal of Personality Assessment, 91*, 166–174 Retrieved from: http://www.sas.upenn.edu/~duckwort/images/Duckworth%20and%20Quinn.pdf.

Eisenberg, N., & Spinrad, T. L. (2004). Emotion-related regulated: Sharpening the definition. *Child Development, 75*(2), 334–339. https://doi.org/10.1111/j.1467-8624.2004.00674.x.

Eisenberg, N., & Sulik, M. J. (2012). Emotion-related self-regulation in children. *Teaching of Psychology, 39*, 77–83. https://doi.org/10.1177/0098628311430172.

Elizur, Y., Somech, L., & Vinokur, A. D. (2016). Effects of parent training on callous-unemotional traits, effortful control, and conduct problems: Mediation by parenting. *Journal of Abnormal Child Psychology,* 1–12. https://doi.org/10.1007/s10802-016-0163-7.

Faul, F., Erdfelder, E., Buchner, A., & Lang, A. (2009). Statistical power analysis using G*Power 3.1: Tests for correlation and regression analyses. *Behaviour Research Methods, 41*, 1149–1160. https://doi.org/10.3758/BRM.41.4.1149.

ForFluency (2012). *Fluency Rater. [Mobile application software].* Retrieved from: http://download.cnet.com/Fluency-Rater/3000-2129_4-75357634.html.

Franken, M. J. (2005). Experimental treatment of early stuttering: A preliminary study. *Journal of Fluency Disorders, 30*, 189–199. https://doi.org/10.1016/j.jfludis.2005.05.002.

Gillam, S., & Gillam, R. (2008). Teaching graduate students to make evidence-based intervention decisions: Application of a seven-step process within an authentic learning context. *Topics in Language Disorders, 28*(3), 212–228. Retrieved from http://catalogue.curtin.edu.au.

Goodman, R. (1997). The strengths and difficulties questionnaire: A research note. *Journal of Child Psychology and Psychiatry, 38*, 581–586. https://doi.org/10.1111/j.1469-7610.1997.tb01545.x.

Goodman, R., Ford, T., Simmons, H., Gatward, R., & Meltzer, H. (2000). Using the Strengths and Difficulties Questionnaire (SDQ) to screen for child psychiatric disorders in a community sample. *British Journal of Psychiatry, 1*(177), 534–539. https://doi.org/10.1192/bjp.177.6.534.

Hamill, S. K. (2003). Resilience and self-efficacy: The importance of efficacy beliefs and coping mechanisms in resilient adolescents. *Colgate University Journal of the Sciences, 35*, 115–146. Retrieved from http://groups.colgate.edu/.

Johnson, K. N., Walden, T. A., Conture, E. G., & Karrass, J. (2010). Spontaneous regulation of emotions in preschool children who stutter: Preliminary findings. *Journal of Speech Language and Hearing Research, 53*(6), 1478–1495. https://doi.org/10.1044/1092-4388(2010/08-0150).

Karrass, J., Walden, T. A., Conture, E. G., Graham, C. G., Arnold, H. S., Hartfield, K. N., et al. (2006). Relation of emotional reactivity and regulation to childhood stuttering. *Journal of Communication Disorders, 39*, 402–423. https://doi.org/10.1016/j.jcomdis.2005.12.004.

Kazdin, A. E., & Blase, S. L. (2011). Rebooting psychotherapy research and practice to reduce the burden of mental illness. *Perspectives on Psychological Science, 6*(1), 21–37. https://doi.org/10.1177/1745691610393527.

Kesebir, S., Duru, G., Kucuksubasi, Y., & Yaylaci, E. (2013). The relation between affective temperament and resilience in depression: A controlled study. *Journal of Affective Disorders, 148*(2-3), 352–356. https://doi.org/10.1016/j.jad.2012.12.023.

Kraft, S. J., Ambrose, N., & Chon, H. (2014). Temperament and environmental contributions to stuttering severity in children: The role of effortful control. *Seminars in Speech and Language, 35*(2), 80–94. https://doi.org/10.1055/s-0034-1371753.

Kraft, S. J., Lowther, E., & Beilby, J. (2018). The role of effortful control in stuttering severity in children: Australian replication study. *American Journal of Speech-Language Pathology,* 1–15.

Langevin, M., Packman, A., & Onslow, M. (2010). Parent perceptions of the impact of stuttering on their preschoolers and themselves. *Journal of Communication Disorders, 43*(5), 407–423. https://doi.org/10.1016/j.jcomdis.2010.05.003.

Manning, W. H., & DiLollo, A. (2018). *Clinical decision making in fluency disorders.* San Diego, CA: Plural Publishing Incorporated.

Masten, A. S. (2001). Ordinary magic. *American Psychologist, 56*(3), 227–238. https://doi.org/10.1037/0003-066X.56.3.227.

Millard, S. K., Edwards, S., & Cook, F. (2009). Parent-child interaction therapy: Adding to the evidence. *Journal of Speech-Language Pathology, 11*(1), 61–76. Retrieved from http://link.library.curtin.edu.au/.

Okoli, C., & Pawlowski, S. D. (2004). The Delphi method as a research tool: An example, design considerations and applications. *Information & Management, 42*(1), 15–29. https://doi.org/10.1016/j.im.2003.11.002.

Pennington, L., & Thomson, K. (2007). It takes two to Talk – The Hanen Program® and families of children with motor disorders: A UK perspective. *Child: Care, Health*

and Development, 33(6), 691–702. https://doi.org/10.1111/j.1365-2214.2007.00800.x.

Plexico, L. W., & Burrus, E. (2012). Coping with a child who stutters: A phenomenological analysis. *Journal of Fluency Disorders, 37*(4), 275–288. https://doi.org/10.1016/j.jfludis.2012.06.002.

Rothbart, M. K., Ahadi, S. A., Hershey, K. L., & Fisher, P. (2001). Investigations of temperament at three to seven years: The Children's Behavior Questionnaire. *Child Development, 72*(5), 1394–1408. https://doi.org/10.1111/1467-8624.00355.

Sanders, M. (2012). Development, evaluation, and multinational dissemination of the Triple P-Positive Parenting Program. *Annual Review of Clinical Psychology, 8*, 345–379. https://doi.org/10.1146/annurev-clinpsy-032511-143104.

Sanders, M. R., Morawska, A., Haslam, D. M., Filus, A., & Fletcher, R. (2014). Parenting and Family Adjustment Scales (PAFAS): Validation of a brief parent-report measure for use in assessment of parenting skills and family relationships. *Child Psychiatry & Human Development, 45*(3), 255–272. https://doi.org/10.1007/s10578-013-0397-3.

Schwenk, K. A., Conture, E. G., & Walden, T. A. (2007). Reaction to background stimulation of preschool children who do and do not stutter. *Journal of Communication Disorders, 40*(2), 129–141. https://doi.org/10.1016/j.jcomdis.2006.06.003.

Shaw, D. S., Owens, E. B., Giovannelli, J., & Winslow, E. B. (2001). Infant and toddler pathways leading to early externalizing disorders. *Journal of the American Academy of Child & Adolescent Psychiatry, 40*(1), 36–43. https://doi.org/10.1097/00004583-200101000-00014.

Speech Pathology Australia (2010). *Code of ethics.* Melbourne, Victoria: Author.

Starkweather, C. W. (1987). *Fluency and stuttering.* Englewood, NJ: Prentice-Hall.

Ungar, M. (2015). *Michael Ungar, PhD.* Retrieved from http://www.michaelungar.com/for-parents.

Vanryckeghem, M., Brutten, G. J., & Hernandez, L. M. (2005). A comparative investigation of the speech-associated attitude of preschool and kindergarten children who do and do not stutter. *Journal of Fluency Disorders, 30*(4), 307–318. https://doi.org/10.1016/j.jfludis.2005.09.003.

World Health Organization (WHO) (2001). *International classification of functioning, disability and health.* Geneva, Switzerland: World Health Organization.

Yaruss, J. S. (2010). Assessing quality of life in stuttering treatment outcomes research. *Journal of Fluency Disorders, 35*, 190–202. https://doi.org/10.1016/j.jfludis.2010.05.010.

Yaruss, J. S., & Quesal, R. W. (2004). Stuttering and the international classification of functioning, disability, and health (ICF): An update. *Journal of Communication Disorders, 37*, 35–52. https://doi.org/10.1016/S0021-9924(03)00052-2.

Yaruss, J. S., Coleman, C., & Hammer, D. (2006). Treating preschool children who stutter: Description and Preliminary evaluation of a family-focused treatment approach. *Language, Speech, and Hearing Services in Schools, 37*, 118–136.

Kerianne C. Druker is a PhD candidate in the School of Occupational Therapy, Social Work and Speech Pathology at Curtin University, Perth, Western Australia. She is a certified practising speech pathologist. Her clinical and research areas of interest include treatment of fluency and speech/language disorders in paediatric populations and examining the role that self-regulation plays in early-onset stuttering.

Trevor G. Mazzucchelli is a registered clinical psychologist who has specialised in parenting interventions for families of children with behaviour disorders. He is also a senior lecturer in clinical psychology in the School of Psychology at Curtin University, Perth, Western Australia.

Janet M. Beilby is an Associate Professor of Stuttering Disorders in the School of Occupational Therapy, Social Work and Speech Pathology at Curtin University, Perth, Western Australia. Her current research interests include parent-administered early treatment initiatives, adolescent stuttering treatment programs and the psycho-social impact of stuttering disorders.

Appendix D

Ethics Application Approval

1. INTRODUCTION

Use this form to apply for an ethical review of human research projects conducted by Curtin University staff or students. On submission your application will be triaged as either low-risk or non-low-risk.

Saving and completing the form:

- Click Save as you progress through the form. We recommend that you click Save every 10-15 minutes.

- To close the form, click the Save button then the Close button. You can return to the form at any time. Do not close the window without saving your work.

- To complete the form, tick the white box in the Complete button then click the Complete button itself.

IMPORTANT: The session timeout period is 30 minutes. You will lose unsaved work if the form is idle for more than 30 minutes. Please click the Save button at regular intervals to save your changes.

Completing the investigators section:

- To add an investigator, click the add ('+') button.

- Search for co-investigators by clicking on the first letter of their surname in the alphabet links that appears at the top of the search page. Type the co-investigator's LAST NAME in the 'Search for a particular entry' text box to narrow your search results. Click on the drop-down arrow next to the 'Select' button to display the results.

- Indicate the Chief Investigator by checking the 'Chief Investigator' checkbox.

- Do not enter start or end dates.

- Select appropriate role for each investigator from the drop down menu.

Please email infoed@curtin.edu.au if you cannot find an investigator in the system.

IMPORTANT NOTE FOR STUDENTS AND CO-INVESTIGATORS: Please add yourself in the investigator section before you close the form. You will NOT have access to the form if you are not listed in the application.

2. INVESTIGATORS

Investigator(Add Investigator)
▼
Investigator

Name
Beilby, Janet

Chief Investigator	Start Date (DD-MM-YY)	End Date (DD-MM-YY)	Role
☑			CI

* Explain how the researcher has sufficient skills and experience to conduct the proposed research (NS 3.3.5).

Dr Janet Beilby is an Associate Professor in the School of Psychology and Speech Pathology and an experienced supervisor of PhD and Honours degree projects.

Dr Beilby has supervised clinical training and research in the Curtin Stuttering Treatment Clinic for over 30 years and her current research interests include parent-administered early treatment initiatives and the psychosocial impact of stuttering disorders across the life-span.

Investigator

Name

Bloch, Kerrianne

Chief Investigator	Start Date (DD-MM-YY)	End Date (DD-MM-YY)	Role
☐			Student

* Explain how the researcher has sufficient skills and experience to conduct the proposed research (NS 3.3.5).

Kerianne completed her Honours project with First Class Honours, in the area of paediatric stuttering disorders. She received an award for her Honours Research in November 2016. She is now completing a PhD under a full scholarship award. She has also been employed as a research assistant at Curtin University since 2015.

Investigator

Name

Hennessey, Neville

Chief Investigator	Start Date (DD-MM-YY)	End Date (DD-MM-YY)	Role
☐			Co-Inv

* Explain how the researcher has sufficient skills and experience to conduct the proposed research (NS 3.3.5).

Dr. Neville Hennessey has a PhD in Psychology and teaches within the speech pathology and psychology programs at Curtin University. He is an experienced supervisor of Doctoral, Masters and Honours students and is also currently the Director of Research Training in the School of Psychology and Speech Pathology. He has supervised research projects in the fields of psycholinguistics and language and speech disorders in children and adults, including stuttering. Neville will contribute to the project as a supervisor and provide support in research methods and data analysis.

Investigator

Name

Mazzucchelli, Trevor

Chief Investigator	Start Date (DD-MM-YY)	End Date (DD-MM-YY)	Role
☐			Co-Inv

* Explain how the researcher has sufficient skills and experience to conduct the proposed research (NS 3.3.5).

Dr. Mazzucchelli is a registered clinical psychologist of 23 years experience who has specialised in parenting interventions for families of children with neurodevelopmental and disruptive behaviour disorders. He is also a senior lecturer in clinical psychology and has conducted and supervised many research projects involving families of children with mental health conditions.

3. GENERAL INFORMATION

* Does the research project have a SCRIPT project ID?

☐ Yes ☑ No

* Indicate the type of project.

PhD

* Has this project been peer reviewed?

Peer reviewed means accepted by a granting body that uses a peer review process (e.g. NH&MRC) or if the project has been approved through the candidacy process at Curtin.

☑Yes ☐No

Attach a copy of the acceptance/candidacy letter to this application.

* Does this research involve any staff going overseas?

☐Yes ☑No

* Does this research involve any students going overseas? (NS4.8.8, NS4.8.18), Curtin WiL

☐Yes ☑No

* List the locations research will be conducted. If the research is being conducted on a Curtin University campus please specify the building and room number/s.

Curtin University Stuttering Treatment Clinic - building 404 - Bentley Campus

Private Speech Pathology clinics:
1. Viva Speech Pathology in Tuart Hill and Joondalup
2. SOS Therapy and Learning Centre, Cloverdale
3. Subiaco Stuttering Clinic

* Provide a lay summary of your project. Include background, aims and hypothesis, methods and anticipated outcomes in your summary.
For assistance in writing in lay language please refer to the Tips for Writing in Plain English.

Background
Children who stutter have been found to have poorer self-regulation skills than their fluent peers. Self-regulation refers to the way one responds to changes in the external environment. Poor self-regulation is a symptom consistent with the attention deficit hyperactivity disorder (ADHD) phenotype, and manifests in disruptive behaviours. Furthermore, a negative relationship has been found between stuttering severity and self-regulation skills, with more severe stuttered speech in children with poorer self-regulation skills. This is significant because the presence of poor self-regulation skills in children who stutter may affect their response to stuttering therapy. The presence of poor self-regulation skills manifesting in disruptive behaviours may compound the already well documented negative psychosocial effects developmental stuttering disorder has in childhood. As such, addressing self-regulation skills may be a crucial component of future stuttering therapy for children.

Aims
This project aims to (a) determine if the presence of disruptive behaviours predicts response to stuttering therapy (b) identify if pre-treatment stuttered speech severity moderates this effect, (c) implement an integrated behavioural and fluency intervention program and determine if this results in improved maintenance of fluency outcomes, as well as improvements in child behavior and parenting practices, and (d) gain qualitative parent evaluations of their experiences with the integrated intervention.

Hypotheses
(a + b) The presence of disruptive behaviours will negatively affect response to therapy, that is, increase number of therapy sessions (Karrass et al., 2006; Kraft et al., 2014; Riley & Riley, 2000). This will be moderated by pre-treatment stuttered speech severity.
(c) The children who stutter with disruptive behaviours who receive the integrated intervention will demonstrate enhanced outcomes when compared to the children who stutter with disruptive behaviours who do not receive the integrated intervention including: reduced social and emotional problems post-treatment and at maintenance, improved parenting practices and confidence post-treatment and at maintenance, and improved fluency outcomes at post-treatment and at maintenance.

Methods
A prospective correlational study will be implemented using existing clinical records to examine the effect of disruptive behaviours on response to therapy. A quasi-experimental design will be implemented in the intervention study. Children who stutter with disruptive behaviours who receive the integrated program will receive fluency therapy in addition to the Group Triple P Program. Fluency, behavioural and parenting outcomes will be obtained. Semi-structured face-to-face qualitative interviews will be conducted with parents of children who receive the integrated intervention at post-treatment.
Analysis
Generalised linear modelling will be used to determine if disruptive behaviours predict response to treatment and to examine the interaction between pre-treatment stuttered speech severity and disruptive behaviours. Multivariate Analysis of Variance will be used to compare behavioural and psychosocial outcomes in children who stutter, following the integrated fluency and behavioural intervention. Analyses of variance will be implemented to compare stuttering severity, child temperament and parenting practices, between intervention groups. All measures will be collected at

pre-treatment, post-treatment and at 3 months follow-up, and comparisons between three intervention groups will be drawn. Qualitative interviews will be transcribed verbatim and thematically analysed using QSR Nvivo 8 qualitative analysis software.

Anticipated Outcomes
The results of this project will ascertain the effect of disruptive behaviours on children's response to stuttering therapy. It will determine if an integrated fluency behavioural intervention is successful in addressing these disruptive behaviours, and improving maintenance of fluency outcomes. An in-depth understanding of parent and child experiences with the integrated intervention will be gained. The results will inform future management of childhood stuttering disorders, support caseload management and prioritisation, as well as strengthen prognostic information available to parents of these children.

* Describe how your research will have an impact on the community.

Poor self-regulation skills in childhood, symptomatic of ADHD and found to be prevalent in childhood stuttering, is predictive of significant long-term negative emotional, social and occupational outcomes (Moffitt et al., 2011). This compounds the potentially negative long-term psychosocial outcomes of living with a stuttering disorder (Beilby, 2014). Addressing these symptoms has been found to alter the negative trajectory for children (Moffitt et al., 2011). The proportion of children who stutter presenting with behavioural self-regulation difficulties and symptoms resembling the ADHD phenotype has not been rigorously investigated to date and remains of significant interest for future stuttering research and clinical practice. Although these children may not meet criterion for ADHD diagnosis, the presence of at least subclinical ADHD symptoms could significantly affect a child's functioning by challenging regulation requirements for aspects of daily living. Therefore, this should be addressed as part of a holistic intervention program to promote positive psychosocial development for these children and their families.
In addition, according to the Demands and Capacities model (Starkweather, 1987) and underscored by previous study findings (Riley & Riley, 1979, 2000), effective management of behavioural volatility will likely facilitate more fluent speech production by reducing emotional reactions that interact negatively with the speech motor system; thus improving capacity to devote resources to maintaining ongoing fluent speech production demands (Karrass et al., 2006).
Therefore, the need to support emotional regulation development in subgroups of children who stutter, with concomitant ADHD symptoms, is underscored by the goal of establishing durable fluent speech production in these children by creating "neural networks for speech motor planning and execution that are not unusually vulnerable to the demands of… emotional state" (Smith & Weber, 2016, p.296), as well as improving psychosocial outcomes for these children and their families (Moffitt et al., 2011).

4. RISK AND MITIGATION

* Outline the potential risks to participants. If potential risks are identified, explain how this research justifies the burden and risk to participants (NS 2.1). ❓

There are no foreseeable risks from participating in this research project.

* If you identified risks in the previous question, outline how you will mitigate the risks identified above and your plan of action for expected adverse events and other identified risks. ❓

No risk.

* Outline the potential harm or risk to researchers. ❓

No risk

* Outline the potential risk to the University and the research. ❓

No risk

5. PARTICIPANT RECRUITMENT AND CONSENT

* Are you recruiting participants?

☑Yes ☐ No

* Does your research involve staff and students from Curtin University?

☐ Yes ☑No

* Describe your target population and sample size. ❓

Target population: children who stutter aged 3-6 years, living in the Perth
Metropolitan area
Total target sample size for the study: 120

Select how you are going to recruit participants (select all that apply) ❓

- ☐   Database/medical records

- ☐   Social media including Facebook, Yammer, LinkedIn, Twitter etc.

- ☐   Classroom or hospital or clinic or community groups etc.

- ☐   Snowball recruitment or word of mouth etc.

- ☑   Print media including flyers, newspapers, newsletters, etc.

* List sources:

Flyers will be sent to all potential participants by the clinic manager

- ☐   Radio/television

- ☐   Other

* Describe your recruitment process.

Participants will be recruited from the Curtin University Stuttering Treatment Clinic
(CSTC) and private community clinics using convenience sampling. The inclusion of
participants from a number of private community clinics in various locations around
Western Australia, in addition to CSTC, aims to strengthen the external validity of
study findings.
Expressions of interest in the form of flyers will be distributed by the clinic manager
at each site. After the parent registers their interest, the clinic manger will distribute
the information sheet and informed consent form (see attachment).
The researcher will not have access to any personal participant details until
participants have provided informed consent (NHMRC, 2007).

* Will participants receive anything in exchange for participating in research? (NS 2.2.10 - 2.2.11) ❓
  ☑ Yes ☐ No

* Detail what the participant will receive, what the value is, and when it will be received.

All children who stutter who participate in the study will be taken off a waitlist to
receive 12 weeks of fluency therapy (each session will cost the standard rate
depending on the clinic that they attend).
Parents in one group will receive the Triple P parenting program for free (a well
researched and successful parenting program). They will have to purchase the
booklet (approx. $30).

Participant consent

* Will participants provide consent? (NS 2.2, NS 2.3) ❓
  ☑ Yes ☐ No

* Describe the process of how you will obtain consent. ❓

Following expression of interest, the participant will be sent an electronic
information sheet and informed consent form by the clinic manager. Information
sheets will be written in plain language, and will include: the aim of the project,
expectation from participants, voluntary participation and ability to withdraw from
the study at any time without penalty. Contact details of the PhD student/researcher
and chief co-investigator will be provided to allow participants to clarify any
questions they may have about the research. The researcher will not have access to
any personal participant details until participants have provided informed consent
(NHMRC, 2007).

* Is there the potential for the participant to be subject to coercion or pressure, including perceived position of power or ❓
people in dependent or unequal relationships? (NS 2.2.9 and NS 4.3)
  ☐ Yes ☑ No

* Does the research use deception, concealment, incomplete disclosure, limited disclosure, an opt-out approach, or ❓
use of information, samples, health information etc., without the specified consent from those persons? (NS 2.3)
  ☑ Yes ☐ No

* Describe the method, why it is essential and how participants will be informed after the study.

The measure - ADHD Rating Scale:Home Version will be renamed to "Behaviour Scale" to avoid biased responses or underreporting from parents due to potential stigma attached to ADHD (Mikami, Chong, Saporito, & Na, 2014).

Commonwealth agencies

* Will you collect or use IDENTIFIED <u>health</u> information held by Commonwealth agencies **without consent**?

☐ Yes ☑ No

Private sector

* Will you collect or use IDENTIFIED <u>health</u> information from the private sector **without consent**?

☐ Yes ☑ No

6. RESEARCH METHODS

* Describe your research methods clearly outlining your study protocol and what each participant will be required to do for the research study (<u>NS 3.1</u> and <u>NS 3.4</u>).
You may want to attach a flow chart or Standard Operating Procedure.

Following recruitment and informed consent, participants will be invited to attend the in-clinic assessment. The in-clinic assessment will occur in a quiet clinic room in the CSTC or the private clinic setting with which the child is familiar , with the following assessments administered by the researcherFollowing recruitment and informed consent, participants will be invited to attend the in-clinic assessment. The in-clinic assessment will occur in a quiet clinic room in the CSTC or the private clinic setting with which the child is familiar , with the following assessments administered by the researcher (total 90 minutes, with a 15 minute break): (a) four measures of speech and language to assess inclusion criteria and (b) Stuttering Severity Instrument-4 (Riley, 2009). To maximise motivation, children will receive a sticker at the end of each of the assessments, as well as a small token reward from a "lucky dip" bag (non-consumable prizes) at the end of the last assessment.
Following assessment and satisfaction of inclusion criteria, parents of participants will be sent the following questionnaires electronically using Qualtrics Survey Software (maximum 1 hour completion time): (1) ADHD Rating Scale-IV: Home Version, renamed "Behaviour Scale", (2) Strengths and Difficulties Questionnaire (Goodman, 1997), (3) The Eyberg Child Behavior Inventory (Eyberg & Pincus, 1999), (4) Child Behaviour Questionnaire (Rothbart et al., 2001) (5) Parenting and Family Adjustment Scale (Sanders et al., 2014) and (6) Emotion Regulation Checklist (Shields & Cicchetti, 1997).
Based on the presence of ADHD symptoms as assessed by parental responses to the ADHD Rating Scale: Home Version, participants will be divided into two groups. One group will comprise children who stutter who present with at least subclinical symptoms of ADHD (Group A), and one group will comprise children who stutter who do not present with these symptoms (Group B).
Children who stutter in Group A (i.e. those with disruptive behaviours) will be randomly allocated to two groups (groups A1 and A2) (see Figure 1). Weekly fluency therapy will commence for all three groups (groups A1, A2, B) for 12 consecutive weeks. In addition to fluency therapy, parents in group A1 will receive the additional Triple P P Parenting Program, comprising five 2-hourly weekly group sessions held at the CSTC (week 2 to week 5 of treatment block; and final session in week 8) as well as three 20-to-30 minute individual weekly telephone consultations with the researcher (who will be a qualified provider of Triple P) starting the week of the fifth group session (week 5 to week 7). To assess treatment effects, as well as maintenance of treatment outcomes, post treatment and follow up quantitative data (3-months post treatment) will be collected from all three groups (A1, A2, B) on the following measures: ADHD Rating Scale: Home Version, SDQ, ECBI, CBQ, PAFAS and SSI-4. For children with previous diagnoses of ADHD, the SSI-4 will be conducted at the same time of day pre and post treatment, and at maintenance, to account for the potential effect of medication on stuttered speech severity (Healey & Reid, 2003). Semi-structured qualitative interviews will be conducted with parents of children who received the integrated behavioural and stuttering management program (Group A1) post treatment, with the aim of eliciting information and gaining an in depth evaluation and qualitative reflections on the effect of the integrated treatment on their children's fluency, behaviour, their parenting practices and confidence, and overall family functioning (see attachment for questions). (total 90 minutes, with a 15 minute break): (a) four measures of speech and language to assess inclusion criteria and (b) Stuttering Severity Instrument-4 (Riley, 2009). To maximise motivation, children will receive a sticker at the end of each of the assessments, as well as a small token non-consumable reward.

* Does your research involve withholding from one group specific treatments or methods of learning, from which they may "benefit" (e.g. in medicine or teaching)?

☑ Yes ☐ No

* Does your research include invasive physical procedures, collection of body fluid, tissue sampling, infliction of pain, psychological interventions, treatments, administration of drugs or other substances or use of a medical intervention

device?

☐ Yes ☑ No

---

* Does your research use medical records where participants can be identified or linked?

☑ Yes ☐ No

---

* Briefly explain your research outcomes and how do you plan to analyse the data?
Provide sufficient detail in this section to describe the data you collect, how you will analyse it and what the outcomes of the research will be.

STUDY 1
Research Outcomes:
Determine if the presence of disruptive behaviours will negatively affect response to therapy, that is, increase number of therapy sessions; and if this is moderated by pre-treatment stuttered speech severity.
In addition, the study will answer the following descriptive research questions:
(a) What is the proportion of disruptive behaviours in children who stutter in Western Australia?
(b) Do children who stutter with disruptive behaviours present differently to clinic than children who stutter without these behaviours on the following demographic variables: participant age, age at onset of developmental stuttering disorder, gender, family history of stuttering; as well as stuttering features including stuttering severity and typography?

Data Analysis:
To determine whether the presence of behavioural symptoms predicts treatment response (i.e., number of therapy sessions) while controlling for potential co-variates, generalised linear mixed modelling (GLMM) will be used. The interaction between disruptive behaviours and pre-treatment stuttered speech severity will be tested using GLMM.
The research questions will be answered by descriptively identifying the proportion of children who stutter who present with disruptive behaviours in relation to the total number of participants. To describe the demographic variables, and pre-treatment stuttering features of both groups, independent sample t-tests will be conducted on the following variables: age (years and months), age at onset of disorder (years and months), time close to onset (weeks between stuttering onset and therapy commencement), stuttered speech severity (mild: 2-5% syllables stuttered (%SS); moderate: 5-10%SS; severe: >10%SS), and typography of stuttering (phrase level, whole-word, or part-word repetitions, prolongations, and blocks). The p value will be adjusted using Bonferroni corrections to reduce the likelihood of a Type I Error. Chi square tests of contingency with Cramer's V effect size will be used to measure between group differences in family history and gender. Any variables that are found to be significantly different between groups will be used as covariates in the hypothesis test.

STUDY 2:
Research Outcomes:
(a) determine if administering a parenting intervention program integrated with stuttering therapy, to address subclinical ADHD behavioural symptoms and stuttered speech in children who stutter, will result in improved fluency and psychosocial outcomes for these children and their parents, and (b) gain an in depth understanding of parents' experience with this integrated intervention.
Data Analysis:
To assess pre-post treatment outcomes, two-way mixed Analyses of Variance (ANOVA) will be conducted. Two ANOVAs will be run for five dependent variables (namely: SDQ, ECBI, PAFAS, ADHD Rating Scale: Home Version, and CBQ); one ANOVA will assess pre-post differences between (i) children who stutter with ADHD symptoms who receive fluency therapy in addition to the additional Triple P component (Group A1) and children who stutter with ADHD symptoms who receive fluency therapy only (Group A2) and the second ANOVA will assess pre-post differences between (ii) children who stutter with ADHD symptoms who receive fluency therapy in addition to the additional Triple P component (Group A1) and children who stutter without ADHD symptoms who receive fluency therapy only (Group B). A third ANOVA will be run when assessing the dependent variable of fluency (measured by the SSI-4), examining differences between Group A2 and Group B. To assess maintenance of treatment outcomes, pre-treatment outcomes will be compared to follow-up treatment outcomes at 3 months post treatment, using similar statistical analyses for each dependent variable.
Study 3 - To gain in-depth understanding of parents' experience with the integrated intervention, qualitative interviews will be thematically analysed using QSR Nvivo 8 qualitative analysis software.

---

* Does your research involve exposing participants to radiation? *Eg. radioisotopes, lasers, x-rays, microwaves, ultra-violet radiation*

☐ Yes ☑ No

---

* Does your research use health information (including biospecimens) that may reveal information that may be important for the health or future health of the donor(s), their blood relatives or their community? (NS 3.4.10, 3.5.1 and 3.5.2)

☐ Yes ☑ No

* Does your research involve human genetics? (NS 3.5)

☐ Yes ☑ No

7. CLINICAL TRIALS

* Is your study a clinical trial? (NS 3.3)   ❓

☑ Yes ☐ No

[All clinical trials need to have institutional approval prior to commencing the study. Please contact ORD-clinicaltrials@curtin.edu.au]

* Are you using any medicine, biological or device not entered in the Australian Register of Therapeutic Goods, including any new formulation of an existing product or any new route of administration; or
A marketed medicine, biological or device beyond the conditions of its marketing approval including new indications extending the use of the product to a new patient group and the extension of doses or duration of treatments outside the approved range?

☐ Yes ☑ No

* Will a placebo/non-treatment group be used? (NS 3.3.10)

☑ Yes ☐ No

* Describe why a placebo or non-treatment group is the best comparator.

In study 2 (i.e. the clinical trial), only one of the three groups of participants will receive the Triple P Program. This is so that the researchers can compare the effect of the program on fluency and behaviour outcomes to other children who stutter who do not receive the additional behavioural component.
If children and parents in the group receiving stuttering therapy and the additional behavioral component have more positive results after therapy than the other groups (e.g. better child behavior, more confident parents, or better fluency outcomes in the children), the participants in the other groups will be offered the additional behavioral component after their first therapy block is completed. ALL three groups will be receiving fluency therapy (i.e. fluency therapy will not be withheld due to implications for early intervention/neuroplasticity).

* Describe the randomisation and blinding process.

Children who stutter will be divided into two groups based on the presence or absence of ADHD symptoms (using the ADHD Rating Scale: Home Version). Then in the group of children who stutter with ADHD, half the children will be randomised to receive the additional parent administered Triple P component, and half will receive fluency only. This randomisation will be done using an envelope.

The researchers will be blinded to each participant's treatment group allocation throughout the study. At pre-treatment data collection, participants will not yet be allocated to treatment group. At post-treatment and follow up data collection points, the only measure to be collected (that is not a parent self-report measure) is the SSI-4. Speech pathologists in private community clinics and student speech pathologists who provide the fluency therapy to each participant will be trained to complete the SSI-4 at these two data collection points. Video-recordings will be made of each participant's post-treatment and follow-up assessment, and cross-checking will occur for reliability testing.

* Has this trial been registered? (NS 3.3.12)

☐ Yes ☑ No

[Please register this trial on a publically accessible register (e.g. ANZCTR) prior to recruitment of participants.]

* Are there arrangements (business, financial or other similar association) between a researcher and supplier of a drug or surgical or other device to be used in the trial? (NS 3.3.4)

No

* Are there any restrictions on publications? (NS 3.3.4)

☐ Yes ☑ No

* Is funding sufficient to conduct and complete the trial as designed? (NS 3.3.2 and 3.3.18)

☑ Yes ☐ No

* Are payments to researcher, participants or the institution likely to influence the design, conduct, findings or publications of the research? (NS 3.3.2 and 3.3.18)

☐ Yes ☑ No

* Are the facilities, expertise and experience available sufficient for the trial to be conducted safely? (NS 3.3.5)

☑ Yes ☐ No

* Does your Participant Information Statement make clear to the participant whether they will have continued access after the trial to treatment they have received during the trial, and on what terms? (NS 3.3.18)

☑ Yes ☐ No

8. PREGNANT WOMEN AND HUMAN FETUS

* Does your research involve gametes and/or embryos?

☐ Yes ☑ No

* Does your research involve women who are pregnant and/or the human fetus?

☐ Yes ☑ No

9. ABORIGINAL AND TORRES STRAIT ISLANDERS

* Does your research involve Aboriginal and Torres Strait Islanders? (NS 4.7)   ❓

☐ Yes ☑ No

10. SPECIFIC PARTICIPANT GROUPS

Children and Young People

* Does your research involve children and young people? (NS 4.2).   ❓

Yes children not considered mature minors

* Address why participation of children or young people is indispensable to this research; and how this study has been designed to be appropriate for children or young people.

The overarching objective of the study is to implement an integrated fluency and parent administered behavioural program for preschool children who stutter. Providing fluency therapy in children as early as possible is efficacious to maximise positive treatment results. Although early intervention is currently effective, there is still approximately 26% of children who do not respond to fluency therapy in isolation. Therefore, we need to use this group of participants (i.e. preschool children) to implement the integrated program to determine if this increases response to therapy for children with disruptive behaviours, who do not respond to fluency therapy alone - to maximise fluency outcomes and alleviate negative long term psychosocial consequences of the disorder.
This study is appropriate as children will be receiving regular fluency therapy for their stuttering disorders (as they would if they were attending regular speech pathology clinics). The additional behavioural component will be a parent training program, where parents will be implementing strategies with their children to improve their self regulation skills - therefore no additional contact between researchers and preschool children will occur for behavioural management.

Working with Children Card details
Refer to the Working with Children Check for Researchers to determine if your research requires staff and/or students to have a working with childrens check.

Name	Issuing state	Application or card number
Janet Maude Beilby		1264290
Kerianne Chloe Bloch		2252264

Highly Dependent on Medical Care

* Does your research involve people highly dependent on medical care who may be unable to give consent? (NS 4.4)
People who are highly dependent on medical care refer to those who may be unable to give consent. This may be people who are patients in the emergency department or intensive care, unconscious people or people in termal care.

☐ Yes ☑ No

Cognitive Impairment, Intellectual Disability, Mental Illness

* Does your research involve people with a cognitive impairment, an intellectual disability, or a mental illness? (NS 4.5)
Refer to the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition: DSM-5, and Tables for the Assessment of Work Related Impairment for Disability Support Pension

☐ Yes ☑ No

Illegal Activities

* Does your research involve people who may be involved in illegal activities? (NS 4.6)   ❓

☐ Yes ☑ No

Research involving participants in other countries

* Does your research involve participants in other countries? (NS 4.8)

☐ Yes ☑ No

Research involving non-english speakers

* Does your research involve participants whose primary language is not English?

☐ Yes ☑ No

11. CONFLICTS OF INTEREST

* Are there any potential conflicts of interest?   ❓

☐ Yes ☑ No

12. DOCUMENTS TO UPLOAD

Attachments can be added when you have saved and/or completed this form. To upload documents:

1. Go back to the main submission screen
2. Click **Add**. A new window will pop open to upload new documents
3. Click **Choose File** to select document to upload
4. Click **Upload**
5. The pop up window will refresh to allow for more uploads

IMPORTANT: Uploaded documents appear in the main submission screen. To see which documents you have uploaded Click **Close** to close the popup window.
Below are a list of documents you may need to add to your application:

- Peer review documents
- Protocol
- Participant information statement and consent form
- Parent information statement and consent form
- Child information statement and assent form
- Questionnaires/survey instruments
- Data Management Plan
- Translations where a language other than English is used
- Recruitment materials
- Approval from the Radiation Safety Officer
- Risk assessment
- Investigator Brochure or Product Information (clinical trials involving drugs)
- SOL Research Integrity Professional Development program certificate (Staff can access certificates from iPerform. Students can take a screen capture of completion in blackboard)
- Curriculum vitae's of investigators
- Approval to access Curtin students and staff for research purposes

Appendix E

Research Data Management Plan

Managing fluency and disruptive behaviours in children who stutter: An integrated behavioural and stuttering treatment program

Supervisor	Janet Beilby
Data Management Plan Edited by	Kerianne Bloch
Modified Date	23/06/2017
Data Management Plan ID	BEILBJ-HS03736
Faculty	Health Sciences

1   Research Project Details

1.1   Research project title

Managing fluency and disruptive behaviours in children who stutter: An integrated behavioural and stuttering treatment program

1.2   Research project summary

Background Children who stutter have been found to have poorer self-regulation skills than their fluent peers. Self-regulation refers to the way one responds to changes in the external environment. Poor self-regulation is a symptom consistent with the attention deficit hyperactivity disorder (ADHD) phenotype, and manifests in disruptive behaviours. Furthermore, a negative relationship has been found between stuttering severity and self-regulation skills, with more severe stuttered speech in children with poorer self-regulation skills. This is significant because the presence of poor self-regulation skills in children who stutter may affect their response to stuttering therapy. The presence of poor self-regulation skills manifesting in disruptive behavioural symptoms may compound the already well documented negative psychosocial effects developmental stuttering disorder has in childhood. As such, addressing self-regulation skills may be a crucial component of future stuttering therapy for children. Aims This project aims to (a) determine if the presence of disruptive behavioural symptoms predicts response to stuttering therapy (b) identify if pre-treatment stuttered speech severity moderates this effect, (c) implement an integrated behavioural and fluency intervention program and determine if this results in improved maintenance of fluency outcomes, as well as improvements in child behavior and parenting practices, and (d) gain a qualitative understanding of parents' experience who receive the integrated intervention. Methods A retrospective correlational study will be implemented using existing clinical records to examine the effect of disruptive behavioural symptoms on response to therapy. A quasi-experimental design will be implemented in the intervention study. Children who stutter with disruptive behavioural symptoms who receive the integrated program will receive fluency therapy in addition to the Group Triple P Program. Fluency, behavioural and parenting outcomes will be obtained. Semi-structured face-to-face qualitative interviews will be conducted with parents of children who receive the integrated intervention at post-treatment. Analysis Generalised linear modelling will be used to determine if disruptive behavioural symptoms predict response to treatment and to examine the interaction between pre-treatment stuttered speech severity and disruptive behavioural symptoms. Multivariate Analysis of Variance will be used to compare behavioural and psychosocial outcomes in children who stutter, following the integrated fluency and behavioural intervention. Analyses of variance will be implemented to compare stuttering severity, parenting practices and temperament between intervention groups. All measures will be collected at pre-treatment, post-treatment and at 3 months follow-up, and comparisons between three intervention groups will be drawn. Qualitative interviews will be transcribed verbatim and thematically analysed using QSR Nvivo 8 qualitative analysis software. Expected Results The results of this project will ascertain the effect of disruptive behavioural symptoms on children's response to stuttering therapy. It will determine if an integrated fluency behavioural intervention is successful in addressing these disruptive behavioural symptoms, and improving maintenance of fluency outcomes. An in-depth understanding of parent and child experiences with the integrated intervention will be gained. The results will inform future management of childhood stuttering disorders, support caseload management and prioritisation, as well as strengthen prognostic information available to parents of these children.

1.3   Keywords
Disruptive behaviour, stuttering, intervention, fluency

2   Research Project Data Details

2.1   Research project data summary

Source of data: standardised assessments of speech and language, parent report questionnaires, stuttering severity ratings using an stuttering rating instrument.

Description of content: Speech and Language skills of children Parenting practices of parents Temperament characteristics of children Behaviours of children

SPSS analysis program willbe used. 2-way mixed AVOVA, MANOVA, and Generalised Linear Modelling will be used analyse data.

2.2 Will the data be identifiable

- Re-identifiable — identifiers have been removed and replaced by a code, but it is possible to re-identify an individual

2.3 Will data, including biospecimens, be sent overseas?

No

2.4 Data organisation and structure

Data will be given a code to allow data to be re-identifiable Data will be stored on a Curtin secure network. The videos of treatment sessions will be digitally recorded and stored on the network as per above.

3   Research Project Data Storage, Retention and Dissemination Details

3.1 Storage arrangements

The data will be stored on a Curtin secure network drive.

3.2 Estimated data storage volume

Approximately 1GB

3.3 Safeguarding measures

Print data will be kept in a locked filing cabinet in Building 400.449. Electronic data stored on Curtin secure network drive

3.4 Retention requirements

7-25 years (Conducting research where the projects involve children [-18 years])

3.5 Collaboration

Members of the research team involved in the project. Print data will be accessed with a securely hidden key to the locked filing cabinet in Building 400.449. Electronic data stored on Curtin secure network drive will be accessed through a password.

3.6 Data dissemination

Hope to publish in peer reviewed journals:

Journal of Fluency Disorders Journal of Speech, Language and Hearing Disorders

3.7   Embargo period

No.