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Elevated attention deficit hyperactivity disorder symptoms in children who stutter

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ABSTRACT

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

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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 Beljsterveldt, & 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 $n = 92$				
	M	*SD*	*M*	*SD*	*Z*	*p*	Cohen's *r*
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**	−.16*	.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.

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