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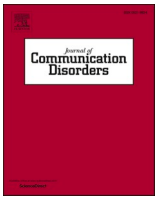

The role of temperament in stuttering frequency and impact in children under 7

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A B S T R A C T

Introduction: Increased emotional reactivity and decreased regulation have been associated with increased stuttering severity and frequency in preschool children who stutter (CWS) and may be predictors for the development of negative reactions to stuttering in young children. Understanding which children are likely to be impacted to a greater or lesser degree has implications for clinical decision making. Associations between temperament and stuttering impact have been explored with older CWS, but not with preschool CWS.
Aim: To investigate the relationship between temperament (specifically emotional reactivity and regulation) and both stuttering frequency and stuttering impact in preschool CWS.
Methods: Data collected at initial assessment for 119 young CWS (age range= 3;00–6;11 years) at a specialist centre for stuttering in London, UK were analysed. The following measures were completed: The Children's Behaviour Questionnaire-Short Form (Putnam & Rothbart, 2006); Palin Parent Rating Scales (Millard & Davis, 2016); The Communication Attitude Test for Preschool and Kindergarten Children Who Stutter (Vanryckeghem & Brutten, 2007); and a stuttering frequency measure.
Results: Emotional reactivity and regulation were not significantly associated with stuttering frequency. Higher scores on negative reactivity were significantly associated with an increased impact of stuttering on the child (from parents' perspective), but not significantly associated with child-reported communication attitude. Positive reactivity was not significantly associated with parent-reported impact of stuttering or child-reported communication attitude. Additional investigation revealed negative affect as a significant predictor of parent-reported impact of stuttering before and after adjusting for age.
Discussion: The results provide evidence to support the role of temperament on the impact that stuttering has in the early years. While the directionality of the relationship between negative reactivity and impact of stuttering is unknown, the importance of targeting emotional reactions in therapy for young CWS is implicated.

1. Introduction

Stuttering is a complex multifactorial neurodevelopmental disorder, with genetic and neurological factors hypothesised as the underlying explanation for stuttering onset (Bloodstein & Bernstein Ratner, 2008). Other influencing factors are thought to include speech motor control, language, emotion and temperament (Ambrose et al., 2015; Walden et al., 2012). In a review of the literature Smith and Weber (2017) concluded that emotion and temperament factors are significant in stuttering in older children and adults but their role in early stuttering requires better understanding and further research.

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1.1. Temperament

Rothbart (2011) defines temperament as "early emotional, motor, and attentional equipment along with the regulating capacities that allow us to control our reactions" (p.6). Temperament is considered to be genetic and neurobiologically (constitutionally) based, evident from early infancy onwards, and relatively stable over time (De Pauw & Mervielde, 2010). DePauw and Mervielde (2010) explain that major elements of temperament are present from early infancy, with other elements being acquired as a result of environmental influences. Rothbart and Bates (2006) describe temperament within three factor level domains: (a) Surgency/ Extraversion ('positive reactivity', i.e., the tendency to show positive emotions such as positive anticipation, low intensity pleasure and smiling/ laughter); (b) Negative Affectivity ('negative reactivity' i.e., the tendency to show negative emotions such as sadness, fear, irritability, discomfort and frustration); and (c) Effortful Control ('emotional regulation' i.e., the ability to regulate emotions, behaviour and attention). Both positive and negative reactivity are reactive temperament domains, beginning to develop early in infancy, with emotional regulation emerging later at approximately two to three years (Rothbart et al., 2007).

There is a variety of methods for assessing temperament in childhood including parent questionnaires and behavioural and psychophysiological methods, all of which have their own advantages (see Rothbart & Bates, 2006). Parent questionnaires are extensively used in temperament research and in clinical practice (Rothbart & Bates, 2006). However, there has been criticism over the validity of parent questionnaires, with some authors suggesting bias in parent responses to questionnaires, including the potential for parents to be influenced by their own temperament, psychopathology, gender bias, and parental expectancy, rather than their child's actual characteristics (Kagan & Fox, 2006; Parade & Leerkes, 2008). The advantage of parent questionnaires compared to physiological testing is that they use the parents' extensive knowledge about their child, and are based on frequent observations of children over a long period of time and in a range of contexts, which cannot be replicated in a laboratory setting (Henderson & Wachs, 2007; Rothbart & Bates, 2006). This long-term perspective over differing contexts seems appropriate for measuring temperament, as temperament is described as relatively stable over time. A further advantage of parent questionnaires is that they are relatively easy to administer, cost effective and easily accessible to clinicians, adding to their clinical utility (Rothbart & Bates, 2006). Rothbart and Bates (2006) also suggest that there is evidence of convergence of questionnaires, laboratory and observational measures of temperament. Thus, while there are some disadvantages to using questionnaires as a method of assessing temperament in preschool children, questionnaires continue to be the most prominent method, providing a useful perspective with established objective validity and clear clinical advantages in terms of accessibility, cost and time efficiency (Rothbart & Bates, 2006).

1.2. Temperament and stuttering

In recent years there has been increasing interest in the relationship between temperament and stuttering in young CWS, with many hypothesising an association (Conture & Walden, 2012; Smith & Weber, 2017). Most of the research in this area has focused on between-group temperamental differences comparing CWS and children who do not stammer (CWNS), with studies also investigating temperament in relation to stuttering frequency and/or severity, and persistence. Empirical studies have used a variety of methods of assessment including parental report, behavioural methods, and psychophysiology. Research findings are varied but in general it is reported that CWS are more emotionally reactive, with increased negative reactivity, reduced emotional and attention regulation, and are less adaptable to change compared to non-stuttering peers (for detailed review see Jones et al., 2014a). Notably, in a large-scale longitudinal study investigating subtypes of persistence and recovery in CWS, Ambrose et al. (2015) reported significantly higher scores in negative reactivity in children whose stuttering persisted, but this was only significant for recovery / persistence during the first year.

1.2.1. Temperament and stuttering frequency and severity

Overall, research investigating the relationship between stuttering severity and/or frequency with temperamental traits in CWS is inconclusive with mixed findings (Alm, 2014). Studies using experimental tasks with CWS under 7 years generally find a relationship with stuttering frequency and/or severity and the temperament domains of emotional reactivity and regulation (Jones et al., 2014b). However, studies using parent-report measures of temperament with this age group report inconsistent findings in these domains (Eggers et al., 2010;Kefalianos et al., 2017; Kraft et al., 2014; 2019; Tumanova et al., 2011).

Jones et al. (2014b) used direct coded behavioural observations to assess the relationship between emotional reactivity and regulation in stuttered and fluent speech in CWS ($n = 8$) and age and gender matched non-stuttering peers ($n = 8$) aged 3;01–5;09. Jones and colleagues reported that within-group analysis revealed significantly increased negative reactivity associated with stuttered compared to fluent speech in CWS, suggesting preschool CWS have reduced ability to regulate their negative emotions. These findings are aligned with previous studies using behavioural and psychophysiological methods reporting reduced regulation as associated with increased stuttering in young CWS (Arnold et al., 2011; Ntourou et al., 2013). Arnold et al. (2011) used behavioural and psychophysiological methods to assess whether stuttering is related to emotional reactivity and regulation in young CWS ($n = 9$, age range= 3;0–5;11) and CWNS ($n = 9$) matched on age (+/- 9 months) and gender. Findings indicated that the decreased use of regulatory strategies was related to increased stuttering in CWS, and the use of more regulatory strategies during narrative resulted in reduced stuttering. Ntourou et al. (2013) used behavioural observation to investigate emotional reactivity (positive and negative) and emotional regulation (self-talk and distraction) in response to a neutral and frustrating experimental task in CWS ($n = 18$) and CWNS ($n = 18$), aged 3;0–5;11. For CWS, increased self-talk during the neutral and frustrating experimental task was associated with increased stuttering in a subsequent narrative task. Further, Choi et al. (2016) reported that increased positive reactivity was associated with increased stuttering in CWS ($N = 47$, aged 3;0–6;11) using psychophysiological methods (skin conductance level); with

increased negative reactivity only associated with increased stuttering in a positive emotion inducing situation.

Kraft et al. (2014) used parent questionnaires to investigate the relationship between parent and clinician reported stuttering severity with temperament (measured using the Child Behaviour Questionnaire, CBQ) and external environments. Findings revealed a significant association only between reduced emotional regulation and increased stuttering severity in CWS ($N = 69$, aged 2;04- 5;09 years). These findings were later replicated with a larger sample size ($N = 98$), using the same data collection methods, and a wider age range of 2;04–12;06 years (Kraft et al., 2019).

Walden et al. (2012) went beyond the primary constructs of temperament and considered the role of temperamental constellations in CWS ($n = 19$, aged 3;1–5;0) and CWNS ($n = 22$, aged 3;0–4;9) using parent questionnaires and behavioural observations. For CWS, Walden and colleagues reported that less stuttering was observed with higher levels of both negative reactivity and emotional regulation. However, increased stuttering was observed in CWS with higher levels of negative reactivity without concomitant regulation. These findings suggest that efficiently regulated emotions facilitate fluent speech in children who have a more reactive temperament.

However, the findings across studies are equivocal, with some studies reporting no significant association between stuttering severity and parent-reported temperament in preschool CWS (Eggers et al., 2010; Kefalianos et al., 2017; Tumanova et al., 2011). Eggers et al. (2010) used the CBQ to investigate temperament in CWS ($n = 58$) and non-stuttering peers ($n = 58$), aged 3;4–8;11 years. Eggers and colleagues found no significant association between parent-reported temperament and clinician measured stuttering severity. Kefalianos et al. (2017), in a large longitudinal community cohort study of preschool CWS ($N = 173$) at ages 3, 4 and 6 years, also found no association between parent-reported temperament (measured using the Short Temperament Scale; Prior et al., 2000) and parent- and clinician-rated stuttering severity. Similarly, Tumanova et al. (2011) assessed the association between parent-rated temperament (using the CBQ) and the duration of sound prolongations in a clinician measured speech sample of CWS ($N = 19$, aged 2;10–5;10), revealing no significant relationship between temperament and length of prolongations.

These varied findings may be explained by methodological differences, including the small sample sizes in some of the studies, the varied age ranges of the included participants, and whether community, clinical or research populations were studied. Additionally, studies investigating stuttering frequency as opposed to severity may not be comparable, and it is likely that variability across studies in the methods used to assess stuttering frequency or severity will account for the variation in findings: some studies used clinician-rated stuttering frequency or severity alone, whereas others used parent-rated either alone or in addition to clinician-rated severity. Similarly, variability in the methods used for examining temperament (i.e., parental questionnaires, behavioural and psychophysiological methods) are also likely to be influential.

It is unclear from current research whether there is an association between stuttering frequency and temperament in preschool CWS. Larger scale studies are required to substantiate the presence of, or absence of, a salient relationship between temperament and stuttering severity and frequency (Kefalianos et al., 2017). The purpose of the present study was to investigate the relationship between emotional reactivity and regulation with stuttering frequency in a large UK clinical cohort. The main advantage of investigating a clinical cohort is that it is a true representation of the population of young CWS who present in a clinic for stuttering treatment. Thus, the findings of this study can be applied by clinicians to the population of children with whom they work. Cohorts that are recruited for research purposes are selected to adhere to particular criteria and consent to participate in research has to be sought, with the result that some will not participate and therefore the sample may be biased. Using data that are routinely collected and recorded as part of clinical practice means that more data from more children are included, since children are not being selected to opt in to additional research activity, and therefore the sample is more representative and inclusive of a clinical caseload.

1.2.2. Temperament and impact of stuttering

The impact of stuttering varies across individuals and while not all young CWS are impacted cognitively, behaviourally, and affectively, there are those who are (Langevin et al., 2010). Based on parent report, there is evidence that preschool CWS close to onset can experience negative emotional and psychological consequences of stuttering, including frustration, anger, sadness, and upset, alongside behavioural responses such as withdrawing, ceasing to talk, or directly commenting on being unable to speak (Boey et al., 2009; Langevin et al., 2010). Peer responses may further contribute to the potential impact of stuttering on young CWS. While some research has shown positive or neutral peer responses to a young child's stuttering (Langevin et al., 2009), young children also experience teasing and exclusion from peers (Ezrati-Vinacour et al., 2001; Langevin et al., 2009).

The child's experience of stuttering, including their own emotional responses and how others respond to them, is likely to impact how they view themselves as communicators. In a meta-analytic review, Guttormsen et al. (2015) reported that preschool CWS consistently showed more negative attitudes toward communication than peers who do not stutter. Awareness and the impact of stuttering increases as children get older with more negative speech attitudes reported in older children and adults who stutter (Blood et al., 2001; DeNil & Brutten, 1991; Vanryckeghem & Brutten, 2011), as well as a high incidence of anxiety related disorders in older CWS and adults who stutter (Iverach et al., 2016; McAllister et al., 2015). In short, stuttering has the potential to negatively impact preschool CWS cognitively, affectively and behaviourally in the early years, with long term consequences.

There is a limited range of methods for assessing stuttering impact from the perspective of the preschool CWS. The Communication Attitude Test for Preschool and Kindergarten Children Who Stutter (KiddyCAT; Vanryckeghem & Brutten, 2007) is an assessment tool developed to investigate one cognitive component of stuttering in young CWS, specifically the child's view of him/herself as a communicator. The Palin Parent Rating Scales (Palin PRS; Millard & Davis, 2016) incorporates questions relating to the behavioural and affective aspects of stuttering impact from a parent perspective.

It has been suggested that temperament might explain some of the individual differences in the sensitivity or response (reactivity) to environmental demands, or in the regulation of reactions (Caprara & Cervone, 2000), and children who experience greater negative

reactivity are at risk for later developing anxiety (Côté et al., 2009). Theoretically it is proposed that young children with a genetic predisposition to stuttering, presenting with temperamental tendencies of increased negative affect, with reduced adaptability to change, and reduced emotional and attentional regulation, may be at increased risk of starting to stutter, with an increased likelihood of developing negative reactions to stuttering, and a reduced resilience to cope long-term (Conture & Walden, 2012; Jones et al., 2014a; Rocha et al., 2019; Walden et al., 2012).

Research investigating the possible association between temperament and impact of stuttering is sparse, with a small number of studies that have been carried out reporting associations in adolescents and adults who stutter (Bleek et al., 2012; Lucey et al., 2019; Stipdonk et al., 2014) and school-age CWS (Eggers et al., 2021). Eggers et al. (2021) investigated the relationship between temperament and stuttering impact using parent- and self-report questionnaires in a UK clinical cohort of school aged CWS ($N = 123$; aged 9;00 −14;10 years). Findings indicated that an increased impact of stuttering, measured by the Overall Assessment of the Speaker's Experience of Stuttering (Yaruss & Quesal, 2016) was associated with higher negative reactivity (from parent- and self-report measures) and lower positive reactivity (self-report only), as measured using the Early Adolescent Temperament Questionnaire-Revised (Ellis & Rothbart, 2001). Therefore, school-aged children with higher levels of positive reactivity and who are therefore more likely to be outgoing, sociable, and confident were less affected by their stuttering. Children who scored higher in negative reactivity and were more likely to be easily frustrated by their speech and to negatively evaluate their speech, scored more highly on a measure of stuttering impact.

Given that temperament is considered to be innate, it is possible that the significant associations between temperament and impact reported by Eggers et al. (2021) may be present in the preschool years. Investigating the relationship between temperament and stuttering impact in young CWS has the potential to further explain the origins of stuttering impact in this population.

1.3. Potential influencing factors

It is likely that the relationship between temperament, stuttering frequency and impact is complex and will be influenced by a number of factors (Rocha et al., 2019). It might be anticipated that as children become older, or have been stuttering for longer, their attitude to communication and the impact of the stuttering would become increasingly more negative as their awareness and exposure to more negative stuttering experiences increases. Age and time since onset (TSO) variables are related, in that older children tend to have been stuttering longer, but they are not interchangeable. Following their meta-analytic review of the existing literature, Guttormsen et al. (2015) concluded that communication attitudes do become increasingly more negative with age. However, in younger children, no association was found between communication attitude and TSO by Winters and Byrd (2021), and Groner et al. (2016) found that in children aged 4 to 5 years communication attitudes of CWS improved, with more positive attitudes associated with longer TSO. Varied findings have been reported in the few studies which include the association between parent-rated stuttering impact and TSO, with some finding an association (Ntourou et al., 2017) and others finding no significant association (Guttormsen et al., 2021).

Parents' experiences of stuttering (themselves or other family members) may also influence their perception of impact on their child. Rocha et al. (2020) reported that where there is a family history of stuttering, parents' ratings of stuttering impact were higher compared to child-rated impact, in some domains (i.e., reactions to stuttering and quality of life) in school-age children. Similarly, Wheeler et al. (2011) found that parents who stutter themselves rated a higher impact on their preschool child. However, Guttormsen et al. (2021) found no significant association between parent-rated impact and a family history of stuttering in preschool CWS.

Evidence regarding the relationship with treatment status and stuttering impact is varied. Millard et al. (2018) found a significant reduction in child-reported communication attitude and parent-rated impact of stuttering on their child post treatment. On the contrary, Guttormsen et al. (2021) found no significant difference in parent perceived impact of stuttering based on treatment status in young CWS (aged 2–5 years). Thus, it follows that treatment status should be considered as a potential confounding variable.

The findings are inconsistent across studies reporting on the relationship between stuttering impact, including communication attitude, and age, TSO, treatment status and a family history of stuttering. As a consequence, age, TSO, treatment status and family history of stuttering should be considered as potential confounding variables in this study.

1.4. Study aims

This study aims to investigate the relationship between temperament and (1) stuttering frequency; and (2) the impact of stuttering in young CWS. Based on the review of the current literature it is hypothesised that children who have temperamental characteristics of greater negative affect and/or lower regulation will experience higher stuttering frequency and greater stuttering impact. In addition, the current study aims to further explore the relationship between temperament and the impact of stuttering when controlling for the potential confounding factors of age, TSO, family history of stuttering and therapy status.

2. Method

2.1. Participants

All children attending a specialist centre for stuttering in London, UK; aged 3;00 to 6;11, who were considered to be stuttering by both their parent(s) and a specialist speech and language therapist (SLT), and whose parent completed the Children's behavior Questionnaire- Short Form (CBQ-SF; Putnam & Rothbart, 2006) were included in the analysis. All participants were care-seeking, in that parents or other professionals identified stuttering and referred them for an assessment of their stuttering. There were no exclusion

criteria. The study included 119 CWS aged between 3;00 to 6;11 years (36 to 83 months, $M = 58.44$ months, $SD = 13.80$, 84 male and 35 females) and their parents/carers. All measures were completed as part of the child's routine initial assessment between October 2012 and June 2020 Table A. shows the demographic data collected for this cohort, with regards to primary language, hearing status, current language levels, and comorbid diagnoses. Receptive and expressive language levels were assessed using four subtests of the Preschool CELF 2 UK (Preschool Clinical Evaluation of Language Fundamentals; Wiig et al., 2006): Sentence Structure, Concepts and Following Directions, Word Structure, and Expressive Vocabulary. Those children who scored more than 1 SD above or below the mean on the two expressive or receptive subtests were considered to be above or below average, respectively. A child was considered to have a speech delay or disorder if this was identified and confirmed by the SLT at the assessment, or if parents reported that they attend speech and language therapy for speech sound delay or disorder.

The study was approved by the Research Ethics Committee of the School of Health Sciences, City University, London.

Table A
Participant demographic data.

Variable	Number of children ($n = 119$, unless stated otherwise)
Gender	
Male	84 (70.6%)
Female	35 (29.4%)
Mean age at time of assessment (months)	58.44 ($SD = 13.80$; range = 36 - 83)
Mean time since onset (TOS) of stuttering (months)	23.4 ($SD = 13.78$, range= 3 – 72)
Previous therapy for stuttering	
Yes	36 (30.3%)
No	83 (69.7%)
Family history of stuttering	
Yes	76 (63.9%)
	43 (36.1%)
No	
Language background	
English first language	66 (55.5%)
Bilingual first language acquisition (BFLA)	24 (20.2%)
English second language acquisition (ESLA)	29 (24.4%
Receptive language level ($n = 93$)	
Within expected range	82 (88.2%)
Above normal range	4 (4.3%)
Below normal range	7 (7.5%)
Expressive language level ($n = 98$)	
Within expected range	85 (86.7%)
Above normal range	7 (7.1%)
Below normal range	6 (6.1%)
Additional diagnosis	
	108 (90.8%)
None	2 (1.7%)
	3 (2.5%)
Autism Spectrum Disorder (ASD)	1 (0.8%)
	1 (0.8%)
ASD suspected	1 (0.8%)
	1 (0.8%)
Cerebral Palsy (CP)	1 (0.8%)
	1 (0.8%)
Dyslexia	
Kabuki Syndrome	
Mast cell activation disorder	
Glycogen Storage Disorder- type 9 (GSD-9)	
ASD and Dyspraxia	
Speech delay/disorder	
	45 (37.8%)
Yes	74 (62.2%)
No	
Hearing difficulties	
Yes	5 (4.2%)
No	114 (95.8%)

2.2. Measures

2.2.1. Temperament

The Children's Behavior Questionnaire- Short Form (CBQ-SF; Putnam & Rothbart, 2006) is a theoretically derived instrument developed to explore temperament traits in children aged 3–7 years. Parents, or primary carers, rate their child on a 7-point Likert scale ranging from 1 ('extremely untrue of your child') to 5 ('extremely true of your child') on a series of 94 situations, with an option of 'not applicable' if the child has not been observed in the situation described. Scale scores are derived from an average of the corresponding items for each scale, with reversal of scores completed for some items. The scale measures 15 primary temperament domains, which fall under three factor level domains: (1) Extraversion/ Surgency; (2) Negative Affect; and (3) Effortful Control. For this study, these 3 factor level dimensions of the CBQ-SF were used in analyses. Overall, the internal consistency of the CBQ-SF is reported as adequate for research purposes, with overall internal consistency ranging from 0.43 to 0.87 (Putnam & Rothbart, 2006).

2.2.2. Parents' perception of impact

The Palin Parent Rating Scales (Palin PRS; Millard & Davis, 2016) was used to measure the impact of stuttering from a parental perspective. The Palin PRS is a standardised online measure (https://www.palinprs.org.uk/secure/pprs_connect.php) with 19 items rated on a 0–10 scale by parents/carers for children up to 14;11 years, resulting in three overall factor scores. Factor 1 is the impact of stuttering on the child; Factor 2 is the severity of stuttering and the impact on the parents; and Factor 3 is parents' knowledge about stuttering and their confidence in supporting their child. Only Factor 1 (Palin PRS 1) was used in this study as a measure of parental/carer perception on the impact of stuttering on their child. Statements include: 'How frustrated is your child with his speech?'; 'How anxious is your child about his speech?'; 'Does your child speak less because of stuttering?'. A lower overall score in Palin PRS 1 indicates that parents perceive the stuttering to be having a more negative impact on the child. Palin PRS 1 has an internal consistency of alpha 0.865, demonstrating the measure's reliability (Millard & Davis, 2016).

2.2.3. Child's attitude to communication

The Communication Attitude Test for Preschool and Kindergarten Children Who Stutter (KiddyCAT; Vanryckeghem & Brutten, 2007) was used to assess the attitudes of CWS towards their own communication. The KiddyCAT is a verbally administered survey consisting of 12 statements where the child is asked to respond with "yes" or "no" to indicate their view of their own speech. Scores can range from 0 to 12, with higher scores indicating a more negative attitude towards speech. The KiddyCAT has an internal reliability of 0.72 Cronbach alpha for CWNS and 0.75 for CWS, indicating the test is a reliable tool for assessing speech attitude in this age group (Vanryckeghem & Brutten, 2007). Factor analysis results indicate that a single factor of 'speech difficulty' underlies the KiddyCAT questionnaire (Clark et al., 2012).

2.2.4. Stuttering frequency

Stuttering frequency (percentage of syllables stuttered,%SS) was measured in line with published guidance for assessing young CWS (Kelman & Nicholas; 2020) and previous research investigating preschool stuttering (Millard et al., 2018). A speech sample was video recorded in clinic while the child interacted with an unknown adult, describing and answering questions about a series of at least ten 'What's Wrong?' stimulus pictures (Speechmark, 2007), to gain a speaking sample of 150 - 500 syllables. These stimulus pictures are intended to challenge the child linguistically to elicit stuttering. The video was then analysed by a specialist SLT trained in measuring stuttering frequency from the same specialist stuttering centre. Sound, syllable, and monosyllabic word repetitions, prolongations, and blocks were counted as stutters (Conture, 2001). Stuttering frequency (%SS) is widely used clinically and in research to assess the outward behaviours of stuttering (Cordes & Ingham, 1994; O'Brian et al., 2004). Inadequate reliability of stuttering frequency is reported for clinicians in different clinical settings (Cordes & Ingham, 1999), however, O'Brian et al. (2004) reported high reliability values for stuttering frequency measures amongst clinicians from stuttering treatment speciality clinics, as in the present study, and research centres. It is acknowledged that a stuttering severity measure can provide a wider and more comprehensive measure of stuttering, as it takes into account not just stuttering frequency but also features that contribute to the perceived severity (i.e., duration, additional tension and struggle behaviours).

When children did not stutter in the specific speech sample, their diagnosis of stuttering was confirmed by either observation of stuttering behaviours by the specialist SLT during other activities, or by parent/carer report and description of stuttering behaviours outside of the clinic setting. Parents/caregivers are accurate in their ability to identify stuttering (Einarsdóttir & Ingham, 2009; Onslow & O'Brian, 2013; Yairi & Ambrose, 2005), and so this method of confirmation was considered valid for children of this age, where stuttering is highly variable.

2.3. Statistical methods

All statistical analyses were performed using the SPSS statistical software package version 25 (IBM Corp., 2017).

Before conducting the main statistical analyses for each research question, distributions of each variable were explored and checked for test assumptions and bias, including normality (Shapiro-Wilk's test), presence of outliers by visual inspection of boxplots, sensitivity analyses and linearity. Pearson's r correlation was used when all parametric test assumptions were met (i.e., CBQ-SF, Palin PRS 1 and KiddyCAT variables). Where test assumptions were not met, Spearman's Rho was used (i.e., stuttering frequency variable). Bonferroni corrections for multiple comparisons were applied to control for Type 1 error, and the alpha level was adjusted to 0.006 for all correlations.

Where a significant correlation was found, multiple linear regression (MLR) analyses were conducted to include potential confounding factors as explanatory variables in the model. Pearson's *r* correlation and point-biserial correlations were conducted to identify which covariates were significantly correlated with the outcome variable (i.e., impact of stuttering) and to be included in the regression model. All test assumptions for MLR analyses were met.

3. Results

Missing values analysis revealed 13.4% ($n = 16$) of KiddyCAT scores and 1.34% ($n = 2$) of stuttering frequency values were missing. All other variables had full data available. A Missing Values Analysis indicated that (Little, 1988) test of Missing Completely at Random (MCAR) was not significant ($x^2 = 25.669$, $DF = 20$, $p = .177$), indicating that the data are likely to be MCAR. Pairwise deletion was used to accommodate any remaining missing data within correlational analyses. There were no missing data in the variables required for the regression analyses.

3.1. Test assumptions

CBQ- SF

Test assumptions for parametric correlational analyses were met, including Shapiro-Wilk's test (Surgency $p = .568$; Negative Affect $p = .213$; Effortful Control $p = .547$). One outlier was identified in the CBQ-SF Effortful Control variable. Sensitivity analysis was applied demonstrating no significant impact for this participant who was therefore retained.

Palin PRS 1

While the distribution of the Palin PRS 1 variable violated the Shapiro-Wilk's assumption of normality ($p = .01$), the absolute z scores of skewness and kurtosis were within the appropriate boundaries ($Z_{Skewness} = -1.75$, $Z_{Kurtosis} = -1.36$), so parametric analyses were conducted between Palin PRS 1 and other variables with normal distributions.

KiddyCAT

The distribution of KiddyCAT scores violated the Shapiro-Wilk's assumption of normality ($p < .001$), however the absolute z scores of skewness and kurtosis were < 3.29 ($Z_{Skewness} = 1.08$, $Z_{Kurtosis} = -1.89$), therefore parametric analyses were conducted using Pearson's *r*.

Stuttering Frequency

Stuttering frequency (%SS) scores ranged from 0 – 47.5%SS (median = 3.2%). Almost 48% had less than 3%SS ($n = 49$), including 10 (8.5%) with 0%SS in the sample of speech recorded. Due to the number of outliers and lack of normal distribution revealed by Shapiro-Wilk's test ($p < .001$), correlations with stuttering frequency were conducted using nonparametric means, i.e., Spearman's Rho.

3.2. Temperament and the impact of stuttering from the parents' perspective

A Pearson's *r* correlation ($n = 119$) revealed no significant correlation between CBQ-SF Surgency or Effortful Control with Palin PRS 1 (see Table B). There was a statistically significant, moderate negative correlation between CBQ-SF Negative Affect and Palin PRS 1, $r(117) = -0.36$, $p < .001$, with Negative Affect explaining 12.9% of the variation in Palin PRS 1 scores (see Table B). This remained significant when using the Bonferroni correction for multiple analyses at the adjusted alpha value = 0.0006. Thus, increased Negative Affect was associated with parents viewing the stuttering as having a greater impact.

3.3. Temperament and the impact of stuttering from the child's perspective

A Pearson's *r* correlation ($n = 103$) revealed no significant correlation between KiddyCAT scores and the CBQ-SF factor domains (see Table B).

3.4. Temperament and stuttering frequency

Spearman's Rho correlation revealed no significant correlation between stuttering frequency (%SS) and the three CBQ-SF factor domains. See Table B for correlation coefficients.

3.5. Temperament and impact of stuttering when accounting for potentially confounding variables

Pearson's *r* correlations and point-biserial correlations were conducted to determine the covariates to be included in the MLR

Table B
Pearson's product-moment and Spearman's Rho[a] correlation coefficients between CBQ-SF factor domains and KiddyCAT, Palin PRS Factor 1 Impact of stuttering, and stuttering frequency (%SS).

CBQ-SF Factor Domain	KiddyCAT	Palin PRS Factor 1 Impact of stuttering	%SS
Surgency	−0.06	.17	.43[a]
Negative Affect	.11	−0.36*	.03[a]
Effortful Control	.07	−0.16	−0.20[a]

*correlation is significant at the 0.006 level (Bonferonni correction applied).
[a] based on Spearman Rho correlation coefficient.

model. All test assumptions were met. If covariates were found to be significantly correlated with parent reported impact of stuttering on the child (Palin PRS 1), they were included in the model. There was a statistically significant, small positive correlation between age and Palin PRS 1 score $r(117) = 0.20$, $p = .26$, with age explaining 4% of the variability in Palin PRS 1 score. There was no significant correlation between Palin PRS 1 with family history of stuttering, previous therapy for stuttering or TSO. Thus, only age was included as a covariate in the MLR model.

3.6. Relationship between Impact of Stuttering (parent perspective), Age and Temperament

A multiple linear regression (MLR) was conducted to determine whether Palin PRS 1 score can be predicted based on CBQ-SF Negative Affect score and Age, by only including CBQ-SF Negative Affect in the regression equation (Model 1), and then adding age (Model 2). The MLR revealed that Negative Affect contributed significantly to the regression model (Model 1), $F(1116) = 17.3$, $p < .001$, and accounted for 12.1% of the variation in Palin PRS 1 score. The addition of age to the model did not lead to a statistically significant increase in variation in Palin PRS 1 score, and explained an additional 0.9% of the variation in Palin PRS 1 score, R^2 of 0.009, $F(1116) = 1.25$, $p = .27$. The full model of Negative Affect and age to predict Palin PRS 1 score (Model 2) was statistically significant, $R^2 = 0.14$, $F(2, 116) = 9.3$, $p<.001$, adjusted $R^2 = 0.12$. Together both predictors explained 13.8% of the variation in Palin PRS 1 score.

In summary, Negative Affect is a significant predictor of parent reported impact of stuttering before and after adjusting for age. Age is not a significant predictor of parent reported impact of stuttering when adjusting for Negative Affect.

4. Discussion

Using clinical data collected at initial assessment, the current study aimed to investigate the relationship between temperament and stuttering frequency, as well as temperament and the impact of stuttering in a UK clinical cohort of preschool CWS. This is the first study to explore the relationship between temperament characteristics and the impact of stuttering in young CWS. The significant association between negative reactivity in children and parent ratings of the impact of stuttering on the child provides support for the consideration of temperament and stuttering impact in both clinical practice and in research with young CWS.

4.1. Temperament and the impact of stuttering

4.1.1. Negative reactivity

It was hypothesised that young CWS who are more negatively reactive (Negative Affect) would experience a greater impact of stuttering, on the basis that those who score high in negative reactivity are more likely to show anger, frustration and be upset, and would therefore be more likely to negatively evaluate their speech (Eggers et al., 2021; Jones et al., 2014a). In part, the findings supported that expectation, with higher scores on negative reactivity found to be significantly associated with an increased impact of stuttering on the young child, as judged by parents. The significant correlation between negative reactivity and parent-reported stuttering impact was expected, given that the temperament domain of negative reactivity encapsulates negative emotional reactions such as sadness, fear, irritability, discomfort, and frustration (Rothbart & Bates, 2006). These emotional responses are often reported by parents when describing their children's reactions to stuttering (Boey et al., 2009; Langevin et al., 2010) and are included within the Palin PRS Factor 1 subscore. Age was not a significant predictor of parent-reported impact of stuttering in this cohort, so it would seem that in the early years, the impact of stuttering does not increase with age. This suggests that negative affect, rather than age, is a predictor of the impact of stuttering. These findings, combined with those of Eggers et al. (2021), indicate that from preschool through to school-age CWS with higher negative reactivity are more likely to be negatively impacted by their experience of stuttering.

In contrast to expectations, there was no significant association between negative reactivity and children's communication attitude. There are few methods of evaluating the impact of stuttering on young children from their own perspective. While recognising that stuttering may impact in many ways, cognitively, affectively and behaviourally, it was argued that a child's attitude to communication (as measured by the KiddyCAT) is one element of the cognitive impact of stuttering. In contrast, the Palin PRS 1 'Impact on the child' component largely consists of statements that relate to the child's affective reactions to the stuttering, i.e. those features that closely relate to the construct of negative affect. Therefore, one explanation for negative reactivity correlating with the parent-reported measure of impact and not the child-reported measure, is that the two measures are evaluating different aspects of impact.

What is not clear from these findings is whether innate temperament characteristics affect the impact of stuttering, or indeed whether the experience of stuttering can affect an individual's temperament profile. Further longitudinal research would be required to understand the direction of the relationship.

4.1.2. Positive reactivity

It was also predicted that higher scores of positive reactivity would be associated with a reduced impact of stuttering. This was based on the findings of Eggers et al. (2021) who found that those who score high in positive reactivity are less likely to negatively evaluate their own speech. However, no such association was found. It may be that the importance of positive reactivity becomes more significant over time, or with age, and may be further explored with longitudinal research.

4.1.3. Emotional regulation

It has been found that emotional regulation moderates the relationship between negative reactivity, attention biases and anxiety

(Lonigan & Vasey, 2009; Susa at al., 2012), and therefore plays an important role in how children manage their emotional reactions. It was therefore also predicted that lower scores in emotional regulation would be associated with a greater impact of stuttering, with those more able to regulate the negative aspects of reactivity might be expected to be less impacted by their stuttering (Jones et al., 2014a). In contrast to expectations, but in line with Eggers et al. (2021), no significant associations were found.

However, it is possible that emotional regulation is important when considered in the context of emotional reactivity, with negative reactivity and emotional regulation together having greater importance with regard to the impact of stuttering, rather than the individual temperament factor domains in isolation. Beyond the stuttering literature, high negative reactivity matters more when coupled with low self-regulation, and less in the context of high regulation (Eisenberg et al., 2000), a relationship proposed by Conture and Walden (2012) and Walden et al. (2012) within the Dual Diathesis-Stressor Model. Future research may be directed at exploring the relevance of differing temperament constellations, such as the benefits of higher reactivity and higher regulation, rather than higher reactivity and lower regulation for instance.

4.2. Temperament and stuttering frequency

While it was hypothesised that there would be a positive association between emotional reactivity and stuttering frequency, and a negative association between emotional regulation and stuttering frequency, the lack of a significant association between temperament and stuttering frequency is in line with findings from others who have used the CBQ and objective stuttering measures (Eggers et al., 2010; Kefalianos et al., 2017; Tumanova et al., 2011). Other studies have found positive correlations between temperament and both stuttering frequency and severity (Arnold et al., 2011; Choi et al., 2013; 2016; Jones et al., 2014b; Kraft et al., 2014; 2019; Ntourou et al., 2013; Walden et al., 2012). Studies differ with regard to: cohort sizes; age range of participants; whether clinical populations or community cohorts who have not necessarily been referred for support; the use of stuttering severity versus stuttering frequency; speech sample analysis versus parent- or therapist-ratings of severity; and, methods of examining temperament. These many methodological differences make comparisons across studies difficult and there is no clear pattern in the range of methods employed that can explain the differences in findings. In fact, the opposite is true with studies with very different methods yielding similar findings (e.g., Kefalianos et al., 2017; Tumanova et al., 2011).

4.3. Clinical implications

This is a large clinical cohort study, meaning that the findings can be generalised to those who present at Speech and Language Therapy services. Exploring this subset of children means that the findings can be utilised by clinicians to identify children who present at clinic who may be at greater risk of developing more negative attitudes to their communication and more negative emotional reactions to their stuttering.

The association between negative reactivity and impact is important clinically. Children with higher levels of negative reactivity are more likely to be affected by their stuttering and higher levels of negative reactivity have also been found to indicate greater risk of developing anxiety in later years (Côté et al., 2009). Therefore, identifying and prioritising these children for intervention has the potential to reduce the likelihood that social anxiety will reach the elevated levels reported in the literature (Iverach et al., 2016; McAllister et al., 2015). Considering the child's temperamental profile and supporting the parents and child to enhance strategies to reduce and manage negative reactivity has already been proposed by some as an important feature of therapy with children who stutter (Eggers et al., 2012; Franken & Putker-de Bruijn, 2007; Kelly, 2019; Kelman & Nicholas, 2020; Kraft et al., 2019; Yaruss & Reardon-Reeves, 2017). These findings lend further support to those clinical recommendations for young children specifically.

4.4. Limitations

While the use of clinical data ensures that the findings reflect a clinical population and are therefore of clinical relevance, there are inherent limitations. An important issue was one of missing data. Firstly, not all parents who attended the Centre during the period studied had completed the CBQ-SF about their child and were therefore excluded from this study. It is a lengthy questionnaire and, for some, too time-consuming or difficult, meaning that the cohort studied may be biased in a way that has not been considered. Furthermore, there were missing data for other variables including the KiddyCAT score or stuttering frequency measure. Pairwise deletion was applied to maximise the data that could be utilised (Howell, 2007), but other approaches to missing data such as imputation (Sterne et al., 2009) could be considered to increase validity in future studies.

One of the more unusual features of this study was the lack of exclusion criteria for those participants with comorbid diagnoses. The advantage is that the cohort is more representative of the clinical population as a whole, but it is possible that the subpopulations of children with additional learning difficulties, developmental language disorders, speech disorders or autistic spectrum disorder (ASD) could impact the results, if there are certain temperamental traits that are characteristic of those groups. For instance, children with ASD would be expected to exhibit higher levels of reactivity, lower levels of positive reactivity and lower levels of regulation (Chetcuti et al., 2021). However, given the very small numbers of these children in this cohort ($n = 2$), any potential influence on the results is unlikely.

It is important to acknowledge that nearly half of the children in the study had a stuttering frequency of below 3%SS, reflecting an over-representation of very mild stuttering. Having so many children scoring at the lower end of the range could have impacted on the lack of association between stuttering frequency and temperament. It is possible that the data collection methods could have influenced these lower levels, although in this study speech samples were collected in a consistent manner, using collection methods which

were intended to elicit stuttering through a linguistically demanding task, implemented by an unknown adult. Nevertheless, stuttering in young children is highly variable over time and contexts. There is a question of validity with regard to any speech sample obtained and of course this has implications for a study that seeks to explore relationships with a variable that naturally fluctuates so widely. While stuttering frequency data were analysed by specialist therapists trained to carry out these assessments it must be acknowledged that inter and intra-rater reliability was not available for the dataset, thus a lack of reliability data for stuttering frequency is another limitation of this study

Conclusion

This is the first study to explore the relationship between temperament and the impact of stuttering in preschool CWS and the findings lend further support to the role of emotions and temperament in early stuttering. In this cohort there was a significant association found between negative reactivity and a higher overall impact of stuttering, regardless of age, from a parental perspective. Findings suggest that temperament is associated with parents' ratings of impact on the child, rather than child-rated communication attitude or stuttering frequency in young CWS. Given the evidence that children who experience greater negative reactivity are at risk for later developing anxiety, further investigation of this association is warranted. While the directionality of the relationship between negative reactivity and impact of stuttering is unknown, the importance of targeting emotional reactions in therapy for young CWS is implicated.

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Author statement

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