

Temperament and the impact of stuttering in children aged 8-14

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Abstract

Purpose: The goal of this study was to evaluate possible associations between child- and mother-reported temperament, stuttering severity, and child-reported impact of stuttering in school-age children who stutter (CWS).

Method: Participants were 123 CWS (94 boys and 29 girls) who were between 9;0 and 14;10 years old and their mothers. Temperament was assessed with the revised child- and parent version of the Early Adolescent Temperament Questionnaire (EATQ-R; Ellis & Rothbart, 2001). The Overall Assessment of the Speaker's Experience of Stuttering (OASES; Yaruss & Quesal, 2006) was used to evaluate the stuttering impact.

Results: Child- and mother-reported EATQ-R temperament factors correlated moderately. No statistically significant associations were found between temperament and stuttering severity. The temperament factors of Surgency (both child- and mother-reported) and Negative Affect (only child-reported) correlated moderately with the Overall Impact and several subsections (i.e., Speaker's Reactions, Daily Communication, and/or Quality of Life) of the OASES.

Conclusions: More extraverted and less fearful/shy children experience a lower overall impact of their stuttering. Children with higher levels of irritability and frustration experience a higher overall impact of their stuttering. Since children's ratings of temperament were more sensitive to these associations than mothers', this study supports the inclusion of child reported temperament questionnaires in future research.

Keywords: temperament, stuttering, impact, school-age

Introduction

It is widely accepted that stuttering is a neurophysiological, multifactorial condition which is genetic in nature (Smith & Weber, 2017). Stuttering can be associated with increased anxiety, depression and negative perceptions of the self, and people who stutter often report making life choices based on their stuttering. However, how stuttering develops, the severity of the stutter and the impact that it has, varies from person to person, with some experiencing life-inhibiting negative consequences and others barely affected on a day to day basis. This variability is likely to be the result of a complex interaction of intrinsic and extrinsic variables (Starkweather, 2002). Extrinsic variables such as parental support, responses and attitudes of peers and teachers are all likely to affect the overt stuttering and how the stutter impacts on the individual. However, it is also the way and degree to which the person reacts emotionally, cognitively and behaviorally to the stutter itself and their external experiences, which helps explain why, when, how and to what degree the stuttering has an impact. The tendencies to react to stimuli in particular ways and the ability to regulate these reactions are largely explained by temperament. Therefore, this study seeks to explore whether temperament in school aged children who stutter is a factor that may contribute to the experience of stuttering, specifically the impact that the stuttering has on participation and quality of life. Identifying those who are at greater risk of long-term negative consequences of stuttering has implications for service delivery and prioritization of services and identifying factors that may protect against such consequences may inform the development of individual therapy packages.

Temperament, genetically determined (Cloninger et al., 2019; Saudino, 2005) and relatively stable over the lifespan (Bates et al., 2010; Rothbart, 2011), interacts with the individual's internal and external environments, influencing the degree to which those temperamental traits are expressed (Rothbart & Bates, 1998). Temperament influences a

person's interaction with their environment, beginning with children's early adaptations to life (Rothbart, 2011), with "systems of emotional reactivity and attentional regulation influencing the encoding and interpretation of external events, the external cues generated by these events, and the pattern of responses displayed by the person" (Rueda and Rothbart, 2009, p. 20). Temperament influences the child's social emotional development (e.g., Goldsmith, 1996) and individual and family functioning (Rothbart et al., 2000) by modulating learning processes and experiences and the ability to cope with stressors, such as stuttering. This ability develops over time, with temperament influencing the strategies adopted. Therefore, an individual's temperament also affects the degree to which any stressor has an impact on well-being and quality of life (e.g., Lengua, 2003, 2006; Rueda & Rothbart, 2009). Starkweather suggests for instance that a child who is more prone to frustration is more likely to react more quickly or intensely to a stimulus such as dysfluency. This 'frustration response' will be accompanied by physiological reactions which may increase muscle tension, increasing struggle behaviors, stuttering and/or concomitant movements. The tendency to respond with fear may be also be associated with increased stuttering and/or avoidance behaviors. Temperament may therefore provide some explanation for the different reactions and responses to stuttering and the variability in the degree to which stuttering impacts on an individual's participation, emotional state, or general well-being.

Definition of temperament

Temperament has been defined and interpreted differently over the past three decades (Kristal, 2005; Rothbart, 2011, 2015; Strelau, 1998) and different models of childhood temperament have been developed to capture the multiple dimensions of temperament (Mervielde & De Pauw, 2015). Rothbart who takes a psychobiological approach, defines temperament as 'constitutionally based individual differences in reactivity and self-regulation' (Rothbart et al., 2000). The term 'constitutional' refers to the individual's

biological makeup, influenced over time by genetics, maturation and experience. Reactivity refers to the arousability of physiological and sensory response systems. Self-regulation comprises those processes that can modulate (facilitate or inhibit) one's reactivity.

Measurement of temperament

A variety of methods are available for the assessment of temperamental characteristics. Originally questionnaires were used to measure these characteristics (e.g., Carey & McDevitt, 1978; Gartstein & Rothbart, 2003; Walker et al., 2017) but also behavioral observations (e.g. Goldsmith & Rothbart, 1991), interviews (e.g. Garrison, et al., 1990; Biggs, & Williams, 1990) and psychophysical and psychophysiological indicators (Kagan, 1998; McManis et al., 2002) can be used. Temperament questionnaires draw on the extensive and in-depth knowledge of caregivers who have experienced the child's reactions in different situations and over a long period of time (Vaughn et al., 2002).

Rothbart has developed several temperament questionnaires aimed at different age ranges such as the Children's Behavior Questionnaire for children aged 3-7 years (Rothbart, et al., 2001), the Temperament in Middle Childhood Questionnaire (age 7-10); the Early Adolescent Temperament Questionnaire (Capaldi & Rothbart, 1992), and the Adult Temperament Questionnaire (Evans & Rothbart, 2007). The revised version of the Early Adolescent Temperament Questionnaire (EATQ-R; Ellis & Rothbart, 2001) was designed to assess temperament in adolescents aged 9-15 through self-report as well as parent-report questionnaire.

Parents' ratings of temperament

The use of parental temperament questionnaires has a strong empirical basis since a) parents have access to a wide range of child behaviors, b) several caregiver reports showed satisfactory test-retest reliability and cross-time stability (Slabach et al., 1991), and c) several studies have shown a moderate to strong degree of validity for parental reports (Rothbart &

Bates, 1998).

However, there has also been some criticism of the use of parental ratings (Kagan, 1998; Vaughn et al., 2002), with evidence that parental reports may be influenced by a number of subjective, as well as objective, components (Bates & Bayles, 1984; Kitamura, et al., 2015; Majdandzic et al., 2008; Mangelsdorf et al., 2000; Saudino, 2003). Mangelsdorf et al. report that parents' ratings of their child's temperament may be influenced by their own temperament, with mothers who are less extroverted tending to rate children as more difficult than extroverted mothers do. Parents' moods may affect the ratings, with mothers who are depressed rating the children's temperament less accurately. It is also possible that parents may respond in ways that will present their children in more positive, more socially desirable lights. Mangelsdorf et al. go as far as stating that "Given the subjectivity inherent in mothers' ratings, it is clear that reliance on mothers as the sole informant regarding child behavior is not the best methodological approach to the study of child temperament." (P125), although many (e.g., Rothbart, 2011) would disagree with this statement. These biases may help explain findings in some studies that highlight low inter-rater reliability between parent and child ratings of temperament. For example, Capaldi and Rothbart (1992), using the original EATQ, reported agreement levels between mother and child ratings of temperament subscales ranging from $-.05$ through $.70$, averaging $.24$. In a later study of the EATQ, Ellis and Rothbart (2001) identified agreement levels of $.05$ through $.53$, averaging $.40$ (i.e., only mild to moderate correlations, and sometimes no correlations between mother and child ratings of temperament were found).

Following their review of three psychotherapy assessment instruments, Grills and Ollendick (2002) suggested that limited parent-child agreement may be explained by a number of factors, including the child's age, gender and the type of disorder. Factors such as social desirability (on the part of parent or child), family stress/conflict, and parent's own psychopathology may also explain parent-child differences. Clearly, any of these factors

could be relevant and influential when considering temperament and stuttering. Grills and Ollendick suggested therefore to also consider children as valid, reliable informants with regard to their thoughts, behaviors and emotions, and to ideally obtain data from multiple sources, including the perspective of the child.

To date, there is no information about the relationship between parent and child ratings with regard to children who stutter and research in the field has sought only parents' perspectives of temperament. This is unsurprising since the research has focused largely on preschool children.

Temperament and stuttering

Previous research in the area of temperament and stuttering has used questionnaire based, behavioral, as well as psychophysiological measures (Alm, 2014; Ambrose et al., 2015; Eggers et al., 2009, 2018, 2010; Embrechts et al., 2000; Karrass, et al., 2006; Kefalianos et al., 2017; Kraft et al., 2014; Lucey et al., 2019; Choi et al., 2013; Johnson et al., 2010; Ntourou et al. 2013; Choi et al., 2016; Zengin-Bolatkale et al., 2018). Studies comparing stuttering and non-stuttering cohorts of preschool and school-aged children have not yielded consistent findings (see Kefalianos et al., 2012; Jones et al., 2014 for detailed reviews) but generally have shown a) a similar underlying factorial temperament structure for both stuttering and nonstuttering groups (e.g., Eggers et al., 2009), b) increased (negative) reactivity (e.g., Eggers et al., 2010; Jones, et al., 2014) and c) decreased emotional and attentional self-regulation (e.g., Johnson et al., 2010) in people who stutter. Increased reactivity and decreased regulation are likely to have consequences for stuttering severity and impact.

Temperament and the severity of stuttering

There is some evidence that children who stutter experience greater negative affect and

react more intensely to novelty or change in their environment than children who do not stutter (Jones et al., 2014), although findings have not consistently replicated. Nevertheless, stressful events such as stuttering, often provoke fear, anger, or anxiety (e.g., Craig & Tran, 2006). It might be hypothesized therefore that children who stutter may therefore experience these emotions more often than peers who stutter less frequently, linking negative affect to stuttering frequency. Also, in adults who stutter, some evidence has been found that individuals exhibiting greater reactivity to environmental stressors, stutter more severely (Haitani & Kumano, 2016). One might expect that experiencing emotions more intensely and more frequently would also yield higher scores on assessments measuring affective reactions to stuttering and quality of life such as the Overall Assessment of the Speaker's Experience of Stuttering (OASES; Yaruss & Quesal, 2006).

Self-regulation (the ability to manage one's energy, emotions, attention and behavior) has also been associated with stuttering severity. Ntourou et al. (2013) found poor regulation to be associated with increased stuttering. Kraft and colleagues (Kraft et al., 2014; Kraft et al., 2019) conducted two studies investigating stuttering severity and self-regulation as measured using the Children's Behavior Questionnaire in 2-to 12-year-old children. The results of both studies suggested that children exhibiting lower self-regulatory skills have a higher likelihood of displaying more severe stuttering behaviors. Self-regulation does not only play a role in one's emotional responses through the "process of initiating, maintaining, or modulating the occurrence, intensity, or duration of emotional arousal" (Jones et al, 2014, p116), it also relates to attentional functioning (i.e., which is known to play a role in speech-language planning and impact speech disfluencies (e.g., Eichorn et al., 2019).

Yet these associations between reactivity and regulation and stuttering frequency were not found in a study examining temperament and stuttering frequency in adults who stutter (Lucey et al., 2019). This could suggest a developmental change over time. Alternatively, these different findings may be the result of the different populations, with childhood studies

that incorporate preschoolers, by their nature, including children who both persist and recover. Examining the relationship between temperament and stuttering frequency in school aged children may shed some light on this as this population is likely to contain a preponderance of children whose stuttering is persistent.

Temperament and the impact of stuttering

The impact that stuttering has on an individual is also likely to be affected by their temperament and their ability to cope, since the experience of stressors and the ability to cope can directly impact children's and adolescents' well-being (Garcia & Moradi, 2011; Moreira et al., 2015). The ability to cope is dependent upon two processes: a) the automatic reactions to external and internal stressors (reactivity) and b) the volitional regulatory processes intended to modulate the experienced stress (Skinner & Zimmer-Gembeck, 2007). These aversive states generate different possible reaction patterns and coping mechanisms. For instance, when exposed to stressors (which might include stuttering) people who score more highly on negative reactivity are more prone to experience higher levels of frustration, depressive mood and aggression (Ellis, 2002). Coping mechanisms might include: active coping (approach response to change the situation or think more positively about it), avoidant coping (avoid the situation or thinking about it) and support seeking (use of social support to solve the problem).

Lengua and colleagues (2003, 2006; Lengua & Long, 2002) found that higher negative reactivity resulted in a more negative evaluation of stressful situations and increased avoidant coping; positive reactivity was related to a less negative evaluation of stressful situations and more active coping. So, children who stutter who have higher negative reactivity might be expected to avoid speaking situations, have reduced interaction with friends, family and others, and interpret their experiences through a more negative lens. Finally, children's attentional self-regulation predicted more active coping strategies such as direct problem

solving, seeking understanding, and positive cognitive restructuring. Whether children who stutter have greater or lesser attentional control than fluent peers remains unclear (Kefalianos et al., 2012), but may be more prone to the more negative coping styles, resulting in reduced activity and participation. In stuttering therefore, the degree to which a person is able to cope and the degree to which their well-being is impacted will be affected by the extent to which they react to the stuttering and their ability to manage those moments of stress. Temperament may therefore explain, at least in part, why stuttering is experienced, viewed and managed differently by individuals.

Studies in both typically developing children (e.g., Sanmartin et al., 2018) as several patient populations, such as patients with chronic illnesses or obesity (e.g., Hu & Gruber, 2008; Salhofer-Polanyi et al., 2018; Vilhena et al., 2014) have shown that high Surgency and low Negative Affect are associated with lower reported impact scores or with higher quality of life, albeit that not all findings were consistent (e.g., Harper et al., 2014; Laskowska, 2018). Some theoretical conceptualizations about possible underlying mechanisms (e.g., Scheier, & Carver, 1992) hypothesized about how positive reactivity may lead a person to cope more adaptively with stress. Applying these mechanisms to stuttering, leads us to assume that children scoring high on Surgency have a lower likelihood of negatively evaluating (classical conditioning) their moments of stuttering, listener reactions, or stuttering-triggering situations whereas children scoring high on Negative Affect might experience the opposite, resulting in more avoidant coping (Lengua & Long, 2002).

All of the studies investigating temperament in children who stutter have been either solely focused on (e.g., Anderson et al., 2003), or have included (e.g., Eggers et al., 2009, 2010; Kraft et al., 2014), preschool children. However, the relationship between temperament and stuttering may differ between those who persist and those who resolve the stuttering. It is also possible that parents of preschool children who stutter may not be subject to the biases described previously, in the context of a disorder which will, or is expected to, resolve. As

the child gets older and the stuttering increasingly chronic, the relationship between temperament and stuttering may differ to that observed in the preschool population. Therefore, studying an older group of children, without preschool children included, is needed since it will have the advantage of only including children whose stuttering is likely to be chronic.

While there is one study that recently explored the relationship between temperament and the impact of stuttering in adults (Lucey et al, 2019), no studies have evaluated such a possible relationship in school aged children. Lucey et al. did not find any correlations between the major temperament factors and the OASES, only one correlation was found, i.e. higher scores on the frustration scale were related to a reduced general knowledge about stuttering. An earlier study by Bleek et al. (2012) showed that adults with higher negative reactivity and lower positive reactivity were more likely to experience a greater impact of stuttering on their daily life.

Finally, while previous studies have relied on parent-reports, this is the first study that combines parent-reports with self-reports of temperament. Parent forms and evaluations are based on behavioral observation, but a large proportion of children's daily experiences are outside the home, not observed by parents and may differ to what they see at home (e.g., Kagan, 1998).

Aims of the current study

There were three aims of this current study. The first was to evaluate associations between child- and mother-reported temperament, as measured by the EATQ-R. Based on Ellis and Rothbart (2001), one could speculate about positive correlations between similar constructs in the adolescent- and mother-reported versions; however, others have argued that several factors could contribute to a limited parent-child agreement (e.g., Grills & Ollendick, 2002). The second aim was to investigate possible associations between temperament and stuttering

severity, as measured the Stuttering Severity Instrument (SSI-4; Riley, 2009). Based on the earlier described pathways, one would expect higher negative affectivity and lower self-regulation to be related to a higher stuttering severity, however the existing literature in other age groups has been inconclusive (see Eggers et al., 2010; Kraft et al., 2014, 2018; Lucey et al, 2019; Ntourou et al., 2013). The third aim was to investigate possible associations between children's ratings of temperament and the child-reported impact of stuttering, as measured by the EATQ-R and the OASES. Children who score more highly on negative reactivity are more prone to experience higher levels of frustration, depressive mood and aggression. Since these are emotional responses associated with stuttering, it was hypothesized, that higher scores on negative reactivity (in line with findings in adults by Bleek et al., 2012, and Lucey et al., 2019) and lower scores on positive reactivity (in line with Bleek et al., 2012) would correlate with a greater impact of stuttering. In summary, there have been no studies that have explored temperament in a specific target population of early adolescent children who stutter; no studies that have explored temperament as rated from children's perspectives; and, no studies that have explored whether there is a relationship between temperament and stuttering impact in children. This study seeks to investigate these relationships.

Methods

Participants

Participants were 123 English-speaking children (94 boys and 29 girls) who were between 9;0 and 14;10 years old ($M = 11;7$, $SD = 1;07$) and their mothers. Family history information was available for 106 and of these, with 66 (62.3%) reporting a family history of stuttering. Participants were referred to a specialist center for children who stutter in London. All children who attended the center for an evaluation of their stuttering and the factors that influence it completed a battery of assessments at the initial evaluation. Those who attended during a two-year period and who completed the three measures were included in this study

(see Measures section below). Of these, 87 (70.7%) had received therapy in the past. Since it was a clinical population, there were no exclusion criteria.

Details of comorbid conditions are recorded in Table 1. Forty-eight children (39%) were diagnosed with comorbid conditions, 63 (51%) were not, and for 12 children (10%) not all data were available. Only participants with diagnosed conditions are included in the figures. Due to the nature and time taken in the UK for a clinical diagnosis of autism spectrum disorder and Attention deficit hyperactivity disorder, those who were in the process of a formal assessment were also included. With regard to language, unless language needs are previously identified, the majority of children completed the Core Language Subtests on the CELF-5 (or CELF-4). Children under the age of 13 completed Word Classes and Semantic Relationships (receptive language subtests) and Formulating Sentences and Recalling Sentences (expressive language subtests). Children aged 13 and above completed Understanding Spoken Paragraphs and Semantic Relationships (receptive language subtests) and Formulating Sentences and Recalling Sentences (expressive language subtests). Those children who score more than 1SD above or below the mean on the two subtests were considered to be above or below average respectively. Speech disorders which were identified and confirmed at the assessment, or where parents reported having attended speech and language therapy in the past were included.

Approximately a fifth of the current population had language skills below the age expected range, similar to the 20% reported by Arndt & Healey (2001). Twenty percent of the children had a documented history of speech sound disorders (either current or past). These included both articulation errors and phonological disorder and were those that were significant enough to the child or parents to attend speech and language therapy services or be raised as an area of concern during the assessment. The incidence of speech disorders co-existing with stuttering varies amongst studies and these data are not directly comparable for a number of methodological reasons, but this proportion is somewhat less than the figures presented by

Blood, Ridenour, Qualls and Hammer (2003) who reported 33% of children who stutter experiencing articulation errors and 12.7% phonological disorders, from a large cohort of 2,628 children. The proportion of children in this study who had received a diagnosis of ASD or ADHD or were in the process of a formal assessment, is in line with that found by Arndt and Healey (2001). Only two children were diagnosed with tic disorders (1.7%), fewer than those with cerebral palsy. The proportion of children diagnosed with dyslexia was 5%, lower than the suspected rate in both Blood et al. and Arndt and Healey studies, which may be due to differences in services and the ability to access these assessments.

Overall, the population in this study would appear to be broadly similar with regard to their co-existing diagnoses as those in other studies and would appear to be representative therefore of the population which presents at a clinic.

The participants self-identified as having a stutter, were described as stuttering by their parent(s) and this was confirmed by a speech and language therapist specializing in stuttering. Stuttering severity was determined by the SSI-4 (Riley, 2009) using a speech sample of a minimum of 300 syllables during both reading and conversation activities. Sound, syllable and monosyllabic word repetitions, prolongations and blocks were included as stuttered events (Conture, 2001). The average percentage (conversation and reading combined) of stuttered syllables was 9.13 ($SD = 7.11$). Nine participants were classified on the SSI-4 as very mild, 31 as mild, 35 as moderate, 35 as severe, and 13 as very severe. The study was approved by Thomas More's research council. All data were collected at the first visit to the center.

TABLE 1 ABOUT HERE

Measures

Temperament was evaluated by means of the Early Adolescent Temperament

Questionnaire-Revised (EATQ-R; Ellis & Rothbart, 2001). This is a revision of the Early Adolescent Temperament Questionnaire (Capaldi & Rothbart, 1992), a self- and parent-report measure for 9- to 15-year-olds, based on Rothbart's temperament model. Both child and parent versions were administered. The latter were completed by the mothers. The child's version consists of 12 temperament scales clustering under four factors, i.e. Surgency (or positive reactivity), Negative Affect (or negative reactivity), Effortful Control (or self-regulation), and Affiliativeness (see Table 2). Higher scores, respectively point towards a person being more extravert and less fearful/shy (Surgency), more irritable and frustrated (Negative Affect), more able to self-regulate (Effortful Control), or experiencing more pleasure related to closeness to others and low-intensity activities (Affiliativeness). The 65 items are scored on a Likert scale ranging between 1 (almost never true) and 5 (almost always true). The average internal consistency for the instrument is .73 with two scales scoring above .80 (Shyness and Aggression), six scales scoring between .60 and .70 (Activation Control, Affiliation, Frustration, High Intensity Pleasure, Perceptual and Pleasure Sensitivity) and between .65 and .70 for the other scales (Ellis & Rothbart, 2001). The parent's version consists of 62 items clustering under three factors, i.e. Surgency, Negative Affect and Effortful Control. This version does not include the Perceptual Sensitivity and the Pleasure Sensitivity scales because they contain items that would be very difficult for an observer to judge about another person (e.g., "I tend to notice little changes that others do not notice"). The average internal consistency of the parent's version is .73 with two scales scoring above .80 (Affiliation and Inhibitory Control), four scales between .70 and .80 (Frustration, Shyness, Aggression, and Depressive Mood), and the others scoring between .65 and .70.

TABLE 2 ABOUT HERE

The severity of stuttering was measured using the norm-referenced Stuttering Severity Instrument (SSI-4; Riley, 2009). The tool is used to determine the severity of stuttering in terms of frequency, duration, and physical concomitants. Frequency is expressed in percent syllables stuttered and converted to scale scores of 2-18. Duration (average duration of the three longest stuttering moments) is timed to the nearest one tenth of a second and converted to scale scores of 2-18. The four types of physical concomitants, i.e. distracting sounds, facial grimaces, head movements, and movements of the extremities, are scored on a 6-point scale ranging from 0 (none) to 5 (severe and painful looking), converted to scale scores of 0-20. The total score, the sum of all the scale scores, is converted in a severity equivalent ranging from very mild to very severe. Speech samples of reading and conversation with a therapist were video recorded at the initial assessment in clinic and used for the analysis.

The functional impact of stuttering was measured using the OASES (Yaruss & Quesal, 2006). The OASES is an instrument for evaluating stuttering from the viewpoint of the individual who stutters and consists of four sections: General Information (including the child's view of the amount of stuttering, how much they know about stuttering, and perspectives of being a person who stutters); Reactions to Stuttering (including the emotional, physiological and cognitive components of stuttering); Communication in Daily Situations (exploring how hard it is to talk and participate in speaking situations); and Quality of Life (how negatively the child's life is affected and how much the stuttering gets in the way). For children aged 9 to 12, the 60-item school-age version (OASES-S), and for 13- to 15-year-olds the 80-item teenage version (OASES-T) was used. Items are scored on a five-point Likert scale ranging from 1 (not at all) to 5 (completely). The internal consistency for the four sections ranges between .67 and .94 for the OASES-S and between .88 and .98 for the OASES-T (Yaruss & Quesal, 2008). Higher scores reflect a more negative impact of the stuttering.

Data Analyses

The Shapiro-Wilk test was used to test the normality of the distribution of the different participant variables. Correlations between child and parent versions of the EATQ-R were evaluated using Pearson r coefficients. This was done for all temperament factor scales (i.e., Surgency, Negative Affect, Effortful Control, and Affiliation for the child's version; the parent's version does not include the latter factor scale¹). In order to correct for multiple comparisons, a Bonferroni correction (4 child EATQ-R factors and 3 parent EATQ-R factors) was used, resulting in an alpha level of $.05/12 = .0041$.

Correlations between the factor scales of the child and parent versions of the EATQ-R and the SSI-4 scores (Frequency, Duration, Physical Concomitants and overall score) were also evaluated by Pearson r coefficients for normally distributed variables and Spearman Rank coefficients for non-normally distributed variables. The alpha level was adjusted to $.05/16 = .0031$ for the child's version (4 child EATQ-R factors and 4 SSI scores) and to $.05/12 = .0041$ for the parent version (3 parent EATQ-R factors and 4 SSI scores).

Finally, correlations between the factor scales of the child and parent versions of the EATQ-R and the five OASES scores (i.e., General Information, Reactions to Stuttering, Communication in Daily Situations, Quality of Life, and an overall impact score) were evaluated by Pearson r coefficients for normally distributed variables and Spearman Rank coefficients for nonnormally distributed variables. The alpha level was adjusted to $.05/20 = .0025$ for the child's version (4 child EATQ-R factors and 5 OASES scores) and to $.05/15 = .0033$ for the parent version (3 parent EATQ-R factors and 5 OASES scores). Since a

¹ Only clustered factor scales were included in the analyses since it would be incorrect to compare the conglomerate factor score (of 3 scales) for Affiliativeness in the child version with the Affiliativeness scale in the parent version.

substantial group of the children ($n = 48$) had comorbid conditions, the dataset was re-analyzed after this group was removed.

In order to evaluate a possible effect of age on the results, the dataset was also re-analyzed after the participant group was divided by a median split ($Mdn = 136$ months) in two groups, a younger age group between 9;0-11;4y ($n = 59$) and an older age group between 11;4-14;10y ($n = 64$).

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

Results

Table 3 gives an overview of the means and standard deviations for each of the variables.

TABLE 3 ABOUT HERE

Testing of normality

The Shapiro-Wilk test showed that the SSI-4 scores, i.e., Frequency ($p < .001$), Duration ($p < .001$), and Physical Concomitants ($p = .002$) were not normally distributed. Also, the General Information ($p = .01$) and Quality of Life scores ($p < .001$) of the OASES were not normally distributed. Therefore, Spearman Rank coefficients were used to evaluate the associations between the EATQ-R factor scales and these variables.

Correlations between child- and mother-reported EATQ-R

Significant correlations were found between the child and mother EATQ-R factor scores for Surgency ($r = .55, p < .001$), Negative Affect ($r = .38, p < .001$), and Effortful Control ($r = .47, p < .001$). No other significant correlations were found at the adjusted alpha level.

Table 4 provides an overview of all the correlation coefficients. The results did not change when only the children without comorbid conditions were analyzed. Therefore, only the findings for the full dataset were reported. Also, for the youngest age group, results were similar. For the oldest age group however, the correlation between child and mother factor scores for Negative Affect was no longer significant after applying the Bonferroni correction ($r = .32, p = .01$).

TABLE 4 ABOUT HERE

Correlations between EATQ-R and SSI-4 scores

No correlations were detected between the child or mother EATQ-R factor scores and any of the SSI-4 scale scores, i.e., Frequency, Duration, Physical Concomitants or the Overall score (see Table 5). The results for the analyses of the group without comorbid conditions or for either of the two age groups were similar.

TABLE 5 ABOUT HERE

Correlations between EATQ-R and OASES scores

Several significant correlations were found at the adjusted alpha levels between the EATQ-R factors Surgency (both the child and mother versions) and Negative Affect (only the child version) and OASES scores.

Surgency, rated by the child, correlated negatively with Reactions to Stuttering ($r = -.39, p < .001$), Communication in Daily Situations ($r = -.38, p < .001$), Quality of Life ($r = -.40, p < .001$), and Overall Impact ($r = -.42, p < .001$). In other words, higher scores on Surgency resulted in lower impact scores. Surgency, rated by the mother, only correlated negatively with Quality of Life ($r = -.25, p = .002$), and Overall Impact ($r = -.28, p = .002$). Negative

Affect, rated by the child, correlated positively with General Information ($r = .30, p = .002$), Reactions to Stuttering ($r = .34, p < .001$), Quality of Life ($r = .33, p < .001$), and Overall Impact ($r = .35, p < .001$). In other words, higher scores on Negative Affect resulted in higher impact scores. No correlations were detected between Negative Affect, rated by the mother, and any of the OASES scores. See Table 6 for all the correlation coefficients. The data were re-analyzed for only the children without comorbid conditions. Most results remained similar except for the correlations between child-reported Surgency and Communication in Daily Situations ($r = -.20, p = .14$) and child-reported Negative Affect and General Information ($r = .03, p = .83$), Reactions to Stuttering ($r = .22, p = .09$), and Overall Impact ($r = .20, p = .14$).

The results for the re-analyses of the dataset based on age groups are depicted in Table 7. Age seemed to have an effect since different patterns emerged for the younger (less significant correlations) versus the older age group. There was a significant relationship between child ratings of negative affect and OASES scores within each subsection, which was evident in the older group but not the younger group. This suggests that negative affect plays a greater role in the impact that stuttering has as children get older.

TABLE 6 ABOUT HERE

TABLE 7 ABOUT HERE

Discussion

The aim of this study was to examine whether there was a relationship between temperament, stuttering severity and the impact that stuttering has on a clinical population of school-aged children. This is the largest study to date in terms of participant numbers and the first to focus on temperament and stuttering severity in children between 9 and 14 years of

age. It is the first study to examine temperament from the child's perspective and the first to explore the relationship between temperament and stuttering impact. To begin, we investigated whether mother ratings of temperament matched those of children, and then whether levels of Surgency, Negative Affect, Effortful Control, and Affiliativeness (only in child-version) are associated with stuttering frequency and/or stuttering impact. We evaluated associations between child- and mother-reported temperament, and the client-reported impact of stuttering.

Child- versus mother-reported temperament

Since this is the first time that a study about stuttering has reported temperament as rated by the child, the first question examined was whether children and mothers have similar views of the child's temperament. It was considered possible that the nature of stuttering may influence the parental perception of temperament in their child. However, this was not the case. The child and mother ratings of temperament correlated moderately. In general, mothers who rated their children more highly on Surgency, Negative Affect and Effortful Control, had children who rated themselves similarly within those constructs, in other words, the agreement between parents and children found in this study were stronger than sometimes reported elsewhere (Capaldi & Rothbart, 1992; Ellis & Rothbart, 2001). Since many of our participants were early adolescent children, their mothers might still be in a good position to observe their child's behavior, especially infrequent occurring behaviors. As children grow older, this might be more difficult because of a gradual shift of daily activities with parents to activities with peers, away from home. This is in line with earlier findings (Achenbach et al., 1987) that the agreement between parent- and self-report is higher in 6- to 11-year-olds than for 12- to 19-year-olds.

Temperament and stuttering severity

Following a review of the literature, Jones et al. (2014) conclude that temperament in preschool children who stutter is associated with their frequency of stuttering, with children who exhibit strong reactions to novelty and change (i.e., more behaviorally inhibited exhibiting significantly higher frequencies of stuttering. There was no relationship between temperament and stuttering identified in the current study. These findings support those from Eggers et al., (2010) who studied this relationship in children up to the age of 8;11. The only other studies to include children up to the age of 15, as in this study, are those by Kraft et al. (2014 & 2019). In their study of 98 children between the ages of 2;4 and 12;6 recruited from Australia, Kraft et al. (2019) replicated findings of their earlier study (Kraft et al., 2014), which identified a relationship between Effortful Control and stuttering severity. In our view, two main factors may explain the difference between the previous studies and the current findings. The first relates to the ages of the populations studied. The current study focused on children between the ages of 9 and 15 (to coincide with the EATQ-R age parameters), while Kraft and colleagues included children aged 2;4 to 12;6. Related to the age difference is the question of persistence. Jones et al.'s study had very few children where the stuttering would persist and Kraft et al. would have had a mix of children who would persist and recover. The current study, because of the age of the participants, has primarily children where the stuttering persisted. If as Jones et al. suggest, that there is a relationship in preschool children and not in older children as in this study, then the Kraft results may be weighted by the presence of the preschool children in the study, obscuring any non-significance in older children. Effortful control or self-regulation develops with age, with children becoming increasingly able to focus, shift attention and inhibit or activate their behaviors. So, while the association is significant in the early years, as effortful control increases within the population, the relationship is no longer significant in older children, which might suggest that the later developed aspects of effortful control are not related to stuttering frequency.

Furthermore, the fact that self-regulation might be related to stuttering severity in young children but not in older children, might lead us to speculate that other factors influence stuttering severity or are “needed” for stuttering to persist. Individual differences, subtypes, and untapped relations between and among variables may also account for differential findings.

The second point of difference between the studies relates to the measures used. Kraft et al. used the Children’s Behavior Questionnaire (CBQ) to examine temperament, while in the current study the EATQ-R was used for the age range in the current study. Differences between both instruments could have contributed to the different findings. The CBQ has a three-factor structure (Surgency, Negative Affect, and Effortful Control) and the EATQ-R a four-factor structure (Surgency, Negative Affect, Effortful Control, and Affiliativeness) with different factor loadings. The Effortful Control factor consists of 3 scales in the EATQ-R (Activation control, Attention, and Inhibitory control) and of 4 scales in the CBQ (Low intensity pleasure, perceptual sensitivity, Inhibitory control, and Attentional focusing), allowing for a more fine-grained evaluation of Effortful Control. Perhaps most important, the ‘Perceptual sensitivity’-scale clusters under Effortful Control in the CBQ and under Affiliativeness in the EATQ-R, resulting in different scales contributing to Effortful Control in both instruments. On the other hand, the two measures are developed by the same authors and examine the same constructs and might therefore be expected to yield similar findings with regard to temperamental traits. The differing stuttering severity measures may more likely explain the different results. Kraft et al. asked parents to rate the severity of the child’s stuttering on an eight-point severity rating to give an “overall average”, with instructions to “think globally” in order to consider frequency, duration, tension and secondary characteristics. In this current study, speech recordings of reading and monologue were analyzed using the SSI-4 as an evaluation of severity. There are important differences in the severity rating methods employed across the studies, each having their own advantages in

terms of validity. The parent ratings used by Kraft et al. reflect an overview of stuttering over time and across speaking situations, but these are influenced by the parents' observational skills, their own perceptions of what stuttering severity is, their own temperament, and their degree of worry at the time of the rating, and are therefore subject to bias. In contrast, the SSI-4 is structured, with samples elicited under relatively consistent circumstances and analyzed and rated more objectively by a clinician, with less opportunity for subjective bias. However, the SSI-4 analyses reflect stuttering frequency and severity in one circumstance and one time point only, and may not be representative of beyond clinic stuttering or overall struggle. These may therefore have less validity than the parent ratings. Since the methods of the current study and the findings are more aligned to those of Eggers et al., this would add support for the suggestion that the stuttering severity measures may account for the differences. Given the potential benefits of both methods of measurement, future studies may benefit from the inclusion of both.

In general, studies that focus on young children (under 7) alone do find a relationship with stuttering frequency/severity and temperament or emotional arousal/regulation during experimental tasks (Jones et al., 2014). While those that combine young children and school age children exhibit inconsistent findings, the current examination in early adolescent children demonstrates no association. Further, studies in adults who stutter have not found associations between temperament and stuttering frequency (Lucey et al., 2019). Taken in combination, it would seem possible that the importance of temperamental characteristics evident in the preschool years are no longer relevant in relation to stuttering frequency in children whose stuttering is more persistent over the age of 9 years of age. Perhaps somewhat speculative, older children might have become more negatively conditioned and consequently have developed avoidance behaviors, including those yielding an increase in overt fluency (thus decreasing the frequency of stuttered syllables, among other variables, yielding a lower stuttering severity on instruments like the SSI-4.). Thus, Kraft et al.'s use of

a parent eight-point severity rating scale (or another scale), may be an important addition to this (and many other) studies. Longitudinal research would help to explore whether this relationship does indeed change over time and with maturation, which these studies combined would appear to suggest.

Temperament and impact of stuttering

Lucey et al. (2019) looked for associations between the temperament factors Negative Affect, Surgency, and Effortful Control and the OASES in a group of adult people who stutter and found no correlations. The only correlation they found was that increased scores on the subscale of Frustration were associated with reduced general knowledge about stuttering. In the current study in 9- to 14-year-old CWS, we used a highly comparable (but different because of the participants' age difference) Rothbart-based temperament questionnaire, and found several statistically significant correlations. Prior to analysis, it was anticipated that those who exhibit higher levels of Surgency and who therefore are more likely to be outgoing, sociable and confident, would be less affected by their stuttering than those children who are more likely to be shy, reserved and less confident to engage. In contrast, it was predicted that those children who are more easily frustrated would be more affected. Indeed, the findings supported that expectation.

First, the Overall OASES Impact score was negatively correlated with child- and mother-reported Surgency and positively correlated with child-reported Negative Affect. So, on the one hand, more extravert and less fearful/shy children experience a lower overall impact of their stuttering; on the other, children with higher levels of irritability and frustration experience a higher overall impact of their stuttering. These findings are in line with previous studies (e.g., Sanmartin et al., 2018) and seem to validate the hypothesis that higher Surgency and lower Negative Affect scores to help children who stutter cope more easily with stressful events, possibly through a lower likelihood of negatively evaluating their moments of

stuttering, listener reactions, or stuttering-triggering situations.

A similar pattern was observed for the OASES subscores. Those children who reported themselves as having lower Surgency (comprised of High Intensity Pleasure, Shyness and Fear), reported increased Reactions to Stuttering, greater impairment in Communication in Daily Situations, and reduced Quality of Life. Although a similar pattern seemed to emerge with the mothers' ratings of Surgency, after Bonferroni's correction, only the correlation with Quality of Life remained significant. A possible explanation could also be that mothers' perceptions of their children's quality of life, are influenced by their children's Surgency. It might be that, if mothers perceive their children as outgoing and social, they might expect that their quality of life would be greater, regardless of the stuttering, especially since peer interactions and socialization are key in adolescence. Therefore, this study suggests that when exploring children's perceptions of the impact of stuttering, it is important to include the children's ratings of temperament, rather than relying (solely) on parent ratings.

In addition to Surgency levels, adolescent-reported EATQ-R scores yielded further associations, specifically with regard to Negative Affect. Children with higher levels of frustration and irritability, showed less insight into their stuttering and had more negative feelings about their stuttering (General Information), had more Reactions to Stuttering and a more greatly reduced Quality of Life. The 'Reactions to Stuttering' subscore includes questions such as 'how often do you feel helpless/embarrassed/ashamed/frustrated', and it is not surprising that children who consider themselves to be more easily irritated and frustrated would experience these emotions more frequently and keenly in relation to their stuttering. Lucey et al. (2019), in their study of adults, also found a negative correlation with General Information but not with any of the other OASES subscales. So, although our findings differ from those of Lucey et al., they are in line with Bleek et al.'s (2012) findings in adults that

increased neuroticism² was linked to higher self-perceived negative impacts of stuttering across all OASES subscores.

Finally, children's attentional self-regulation predicted more active coping strategies such as direct problem solving, seeking understanding, and positive cognitive restructuring. It was anticipated that low Effortful Control would be associated with increased impact of stuttering. Some aspects of Effortful Control can be 'negative' such as hyper-vigilance or hyper-focus (i.e., inability to disengage one's attention and "move on") or being unable to do something that one wishes to avoid, or inhibiting certain behaviors. This, in turn, might result in less active coping strategies and less positive cognitive restructuring of stressful situations (Lengua, 2003, 2006; Lengua & Long, 2002). Children's ratings of Effortful Control were correlated with the Reactions to Stuttering and the impact on Quality of Life at first analysis, but in contrast to what was predicted, not once the Bonferroni corrections were applied. So, while there was an initial indication that reduced Effortful Control was associated with increased impact, this did not stand up to our stricter analyses.

If we review the results after the group with comorbid conditions were removed, a number of important patterns emerge. These patterns can give us insight into which aspects may be related to stuttering only and which to stuttering in the presence of comorbid problems. This latter group is also extremely important as it makes up a large part of the clinical population with which every stutter therapist works on a daily basis, as discussed earlier. In the group of children diagnosed with only stuttering, we see that higher scores on Surgency, both in the child and maternal version, correlate with a better Quality of Life and a lower overall impact of the stuttering. In addition, we see that higher child-reported scores for Negative Affect correlate with a lower Quality of Life. On the other hand, those correlations that became

² Regardless of difficulties in directly mapping childhood temperament dimensions onto adult personality dimensions, temperament is seen as a precursor of later personality development (Ahadi & Rothbart, 2014). The temperamental dimensions of Surgency and Negative Affect appear to map fairly well on the personality dimensions of respectively Extraversion and Neuroticism (Evans & Rothbart, 2007).

nonsignificant after removal of the comorbid group (e.g., higher Negative Affect correlated with General Information, Reactions to Stuttering, Overall Impact), might have been more related to communication in general and less specific to stuttering.

Age-related differences

In our participant group, age did not seem to have a major effect on the correlation between child- and mother-reported temperament since the correlations for both Surgency and Effortful Control between the child- and mother-reported version were present in both age groups. While the nonsignificant correlation for Negative Affect in the older group might be attributed to the earlier discussed finding that parent-child agreement is higher in younger versus older children (Achenbach et al., 1987), it could simply be the result of the Bonferroni correction. While age did not have an effect on the correlation between temperament and stuttering severity, it did have a major effect on the correlations between temperament and the impact of the disorder. For the oldest age group (11;4-14;10) nine correlations were found between Surgency or Negative Affect and the OASES scales and only three for the youngest group. This was even more apparent for the Negative Affect factor. For the older age group, Negative Affect correlated positively with every OASES scale while no correlations were found in the younger age group. Specifically, higher irritability levels were linked to greater stuttering impact on all subscales for only the older children. This is similar for the mother-reported temperament, albeit to a lesser degree. Only children in the older age group are perceived by the mother as outgoing and sociable, related to a lower overall impact of the disorder and less impaired daily communication.

Implications for treatment

It may be possible to support children who are at greater risk of being more affected by their stuttering as a result of their (lower) levels of Surgency, through therapy. Children who are more fearful can be supported to understand and manage their worries and fears through

cognitive behavior therapy (Stallard, 2019; Kelman & Wheeler, 2015). Children who are more shy and reticent to engage can learn about the social skills that facilitate communication, and develop confidence to communicate (Reardon Reeves & Yaruss, 2013; Cook & Botterill, 2005). Parents can have a role in encouraging communication in the home perhaps by facilitating turn taking, reinforcing participation and helping develop the adolescent's confidence generally. The young person might attempt and persevere with communication as they develop their independence, problem solving skills, resilience and ability to think flexibly, all of which may be a focus in therapy.

Effortful Control is comprised of activation control, attention focus and shifting, and inhibitory control, each of which can be addressed in therapy. A number of researchers and clinicians have advocated for the need to identify the presence of emotional regulation differences in preschool children who stutter, so that therapy can be adapted to meet those individual needs (Eggers et al., 2012a, 2017; Kraft et al., 2019; Kelman & Nicholas, 2020). For school aged children, Solution Focused Brief Therapy enhances the ability to focus and attend to strengths and 'what is going well' (Nicholas, 2015) and elements of mindfulness may also be employed to understand how we attend and respond to thoughts (Harley, 2015). Manning (2010) and others discuss how these cognitive-based approaches assist children and adults identify and use their resources to approach stressful situations with more effective self-regulatory strategies.

Given that the EATQ-R is comprised of behavioral indices, it is possible that the ratings of temperament are influenced by the child's response to the stuttering itself and it is also possible that these may alter perhaps in response to therapy. For instance, if a child is not avoiding speaking as much, is engaging more in social interaction, and is contributing more in speaking situations, it might be anticipated that they would be rated as less shy than previously. If they are less frustrated and irritated by their stuttering, it is possible that they would have improved scores for Negative Affect. In other words, the behavioral changes

resulting from therapy may result in different ratings on an observational measure of temperament such as the EATQ-R. For instance, an outgoing child who was avoiding social situations and not “behaving” in an outgoing manner due to stuttering, might become “outgoing” when able to better manage stuttering and its impact. If children and their parents’ ratings of temperament are influenced by reactions to stuttering, including other measures, such as behavioral and/or psychophysiological measures in research will be important. Exploring temperament longitudinally would be helpful to understand temperament and its relationship with stuttering more fully.

Limitations

Correlational analysis does not enable the direction of any association to be established, so strictly speaking it is not possible to determine whether the temperamental trait results in, or is the result of, the child’s reactions to stuttering, ability to communicate, or quality of life. But, if temperament is considered to be innate and constant over the lifespan, it makes sense that the temperament underpins the reactions to stuttering, the ability to communicate and quality of life, rather than the other way round.

It is important to recognize that the data used in this study were obtained during clinical assessment procedures and analyzed retrospectively. As is typical in clinical datasets, there were data that were missing. This meant that the number of children included in the final study was less than the total number who attended the clinic and it is possible that this was not random. In other words, it is possible that those children/mothers who did not complete the full set of questionnaires required for inclusion in this study, are a particular subset of the population who are now omitted. It is impossible to establish whether the missing data are random or not. However, it could be argued that prospective research may also have failed to capture these children and parents if they are the ones who are unlikely to complete data collection procedures.

It is important to recognize the limits of the external validity of this study. While this cohort would appear to be largely similar to other clinical populations studied, with regard to the presence of additional needs, the population in this study is a clinical population and thus not necessarily representative of the wider population of children who stutter, i.e. those who do not seek therapy. It would be assumed that the children who present at clinic for an assessment and are seeking therapy, are being impacted by their stuttering in some way. If it is not a problem for them, then they would not attend. Therefore, this study is highly likely to be missing a subset of children who would score very low on the impact measures and insight into their temperament is likely to be under-represented. Future research into this area should include a non-clinical population as well as those seeking support.

Conclusions

This is the largest study to date exploring the relationship between temperament and stuttering, the first to include adolescent ratings of temperament, and the first to examine temperament and stuttering impact in adolescent children. There was no relationship identified between adolescent temperament and stuttering severity. The study sheds light on potential factors that may explain contrasting findings to date and highlights the need to examine the temperament of adolescent children separately to those of younger children. The impact of stuttering was greater for children who were lower in Surgency and higher in Negative Affect. The relationship with Effortful Control was no longer significant following corrections for multiple comparisons. Children's ratings of temperament were more sensitive to these associations than mothers' further supporting the need to include children's self-evaluations of temperament in future research.

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