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Abstract

Purpose: The purpose of this study was to investigate whether preschool-age children who stutter (CWS) were more likely to exhibit a temperamental trait of behavioral inhibition (BI), a correlate of shyness, than their peers who do not stutter (CWNS), and whether this temperamental trait affected preschool-age children's speech fluency and language complexity during a conversation with an unfamiliar adult.

Method: Sixty-eight preschool-age children (31 CWS; 37 CWNS) participated. The degree of BI was assessed by measuring the latency to their 6th spontaneous comment and the number of all spontaneous comments during a conversation with an unfamiliar examiner (following Kagan et al., 1987 methodology). Parent report of shyness from the Children's Behavior Questionnaire (CBQ) served as an indirect measure of BI. Children's language complexity was assessed by measuring their mean length of utterance and the number of words spoken. For CWS, the frequency of stuttering and the negative impact of stuttering were also assessed.

Results: First, we found no between-group differences in the degree of BI across the behavioral observation measures. However, CWS were rated shyer by parents than CWNS. Second, for CWS only, higher BI was associated with less complex utterances and fewer words spoken. Third, for CWS, higher BI was associated with fewer stuttered disfluencies produced.

Conclusions: This study provides empirical evidence that behavioral inhibition to the unfamiliar may have salience for childhood stuttering as it affected the quantity and quality of language spoken with an unfamiliar adult. Clinical implications of high BI for the assessment and treatment of preschool-age stuttering are discussed.

Defined as constitutionally-based individual differences in emotional, motor and attentional reactivity and self-regulation that are stable over time (Rothbart & Bates, 1998; 2006), temperament is often viewed as a risk or protective factor for developmental outcomes (Harrison & McLeod, 2010; Rothbart & Bates, 2006). For example, a growing body of literature has revealed associations between temperamental traits and language development in very young children (Dixon & Shore, 1997, Dixon & Smith, 2000, Kubicek et al., 2001, Morales et al., 2000, Mundy & Gomes, 1998, Sally & Dixon, 2007; Slomkowski et al., 1992; Garello, Viterbori, & Usai, 2012; Usai, Garello & Viterbori, 2009). The role of temperament as an important contributor to stuttering development in preschool-age children has been included within a multifactorial perspective on stuttering (Conture & Walden, 2012; Smith & Weber, 2017) and received empirical support from a number of research studies (Ambrose, Yairi, Loucks, & Seery, 2015; Kefalianos, Onslow, Block, Menzies, Reilly, 2012; Seery, Watkins, Mangelsdorf, Shigeto, 2007). However, the specific mechanisms underlying the association between temperament and stuttering are still not well understood. The purpose of this study was to investigate whether preschool-age children who stutter (CWS) were more likely to exhibit a temperamental trait of behavioral inhibition, a correlate of shyness, than their peers who do not stutter (CWNS), and whether this temperamental trait affected preschool-age children's speech fluency and language complexity. Here we present a review of research on the public's perception of temperamental qualities of CWS, and empirical findings from studies that examined this temperamental quality in CWS. Further, we consider the role of the behavioral inhibition trait in regard to stuttering development, leading to our research questions and hypotheses.

One temperamental quality that the public often attributes to children and adults who stutter is shyness. For example, Craig, Tran and Craig (2003) found that a large number of

people believed that adults who stutter are shy, self-conscious and withdrawn. Similar characteristics, such as being guarded, shy, and withdrawn, were attributed to preschool and kindergarten CWS by university students (Betz, Blood, & Blood, 2008) as well as by school-age children (Frank et al., 2003; Hartford & Leahy, 2007). Notably, these findings have been replicated among professionals including both teachers (Lass et al., 1992) and speech–language pathologists (Lass et al., 1989, Silverman, 1982, Turnbaugh et al., 1979, Yairi & Williams, 1970).

Although shyness has been widely attributed to children who stutter, research investigating the personality traits in children who stutter offers somewhat conflicting evidence. Using a parent-report questionnaire in a prospective study of 1,619 children before the onset of stuttering, Reilly et al., (2009) reported that at the age of two, there was no difference in the degree of shyness between those children who subsequently developed stuttering and those who did not. In a follow up study using a parent-report screening tool for behavioral, emotional and social development, the *Strengths and Difficulties Questionnaire* (SDQ; Goodman, 1997), Reilly et al. (2013) reported that four year-old CWS were not different in their behavior and temperament from peers who did not stutter. In line with the results of Reilly et al., (2009, 2013), two additional studies (Eggers et al., 2010; Embrechts et al., 2000) reported no significant difference in shyness between CWS and CWNS measured by a parent-report assessment of temperament, the *Children's Behavior Questionnaire* (CBQ; Putnam & Rothbart, 2006).

In contrast to the above research findings, Anderson, Pellowski, Conture, and Kelly (2003) found that preschool-age CWS scored higher on the adaptability dimension of the *Behavior Style Questionnaire* (BSQ; McDevitt & Carey, 1978), suggesting that they may be slower to adapt to new situations or people. More recently, two large scale United Kingdom and

United States population studies using the SDQ found that preschool-age children who stutter were rated by their parents as more likely to have worries and be unhappy and have difficulties with emotions (Briley, O'Brien, & Ellis, 2019; McAllister, 2016). Although these two large-scale studies did not specifically measure the degree of shyness, results from the SDQ in both studies implied some differences in approach/withdrawal in social situations between CWS and CWNS. These inconclusive findings warrant more investigation into the potential association between shyness and stuttering in early childhood. It may be that a single measure of shyness does not capture the temperamental differences that may be associated with childhood stuttering. Thus, rather than relying on only one tool (most studies to date have used parent-report questionnaires), the assessment of temperament using multiple perspectives, as suggested by Kagan (2007), may be necessary to yield a more comprehensive understanding of the association between shyness and stuttering in early childhood. As reviewed in the sections below, this temperamental trait could have important implications for children who stutter.

Behavioral Inhibition (BI). One of the well-studied temperamental qualities that influences how children approach (or withdraw) from *new* people, situations, and life events is behavioral inhibition (BI). BI, as proposed in the widely accepted theory by Kagan and colleagues (Kagan, 1994; Kagan, Reznick, & Snidman, 1987), is a physiological mechanism that controls the experience of anxiety in response to novel objects, potentially threatening stimuli, and novel situations. Often described as being shy, behaviorally inhibited children are hyper-vigilant to their environments and prone to exhibit a heightened sensitivity to novelty (Kagan, Reznick, & Snidman, 1987), particularly if it is social in nature (Biederman et al., 2001).

The trait of BI can be observed as early as infancy and is reported to be one of the most stable temperamental characteristics in childhood (e.g., Fox, Henderson, Marshall, Nichols, and

Ghera, 2005; Schwartz et al., 2012). Children showing BI tend to express avoidance, distress, or subdued activity when exposed to the unfamiliar; they tend to stop playing and withdraw when around unfamiliar people and are very vigilant of their surroundings during unfamiliar situations. Psychological research of this trait has indicated that this distinct reaction to novelty is driven by the excitability of neural circuits of the limbic system, particularly by increased activity of the amygdala (Barros-Loscertales et al., 2006; Perez-Edgar et al., 2007; Schwartz et al., 2003, 2012). This trait has also been associated with individual susceptibility to negative emotional states (Kagan & Snidman, 1991; 1999) and enhanced negative emotional reactions to naturally occurring stressful life events (Gable et al., 2000). Because the BI trait is associated with an increase in arousal to novel stimuli and heightened emotional responses to unpleasant cues, children who exhibit BI may experience heightened emotional arousal in a situation or an environment that presents a challenge. Research suggests that CWS have an increased risk of the social and psychological impacts related to difficulties with interpersonal communication (Iverach et al., 2011). Starting in the preschool years, these difficulties may be accompanied by negative peer responses (Langevin, Packman, & Onslow, 2010). Thus, those CWS who are behaviorally inhibited may be at a greater risk of the adverse impacts of stuttering. Behavioral inhibition in childhood has also been included as an etiological risk factor for the development of social anxiety disorder in adolescence and adulthood (Wong & Rapee, 2016). This association holds particular significance for childhood stuttering, as this disorder is often accompanied by social anxiety in both adults and adolescents who stutter (Gunn et al., 2014; Iverach et al., 2016; also see Iverach, Rapee, Wong, & Lowe, 2017).

The role of BI for speech and language. The BI trait has also been shown to influence language development in young children. Slomkowski et al. (1992) found that 2-year-olds

characterized as showing high interest in people, high cooperativeness and happiness, and low fearfulness (all consistent with the low BI profile) exhibited higher standardized language scores at age 7. In addition, Usai, Garello and Viterbori (2009) reported that children with the “inhibited” profile had a smaller vocabulary than children with the “uninhibited” profile in their sample of 106 typically developing 28-month-old children. In a follow-up study, Garello, Viterbori, and Usai (2012) examined the relationship between temperament and language ability in 109 twenty-four- to 30-month-old children. Consistent with their previous findings, they reported that the “inhibited” children showed poorer lexical and morphological abilities and a more immature vocabulary compared to the uninhibited group.

As behavioral inhibition has been linked to socialization and coping strategies (Rothbart & Bates, 2006), some proposed that this temperamental characteristic could influence language development through social interactions (Salley & Dixon, 2007). Accordingly, higher levels of shyness in situations involving novelty were strongly associated with impairments in both social motivation and social communication in a study of 3- and 4 year-old children (Salley, Miller, & Bell, 2013). The authors concluded that children who are easy going and relatively socially skilled are better able to establish social relationships, which in turn can promote language development. Conversely, fear of novelty can inhibit social relationships and lead to fewer opportunities for language learning. This hypothesized association may have a special importance for preschool-age CWS. Specifically, preschool-age CWS who are also behaviorally inhibited may be less able to cope with the difficulties in social interactions that result from stuttering. For these children, the adverse consequences of stuttering could be far reaching, affecting not only their social interactions, but also their language development. Related to this hypothesized association, we recently reported that preschool-age CWS who exhibited a stronger

reaction to their own stuttering (as rated by their parents on a standardized questionnaire from the Test of Childhood Stuttering) demonstrated reduced utterance length and complexity during a conversation with an unfamiliar examiner (Tumanova, Choi, Conture, & Walden, 2018). Although the effects of children's age and language ability were controlled in the study, behavioral inhibition was not specifically assessed, leaving the possibility that the reported association could be impacted by children's temperament.

Given the significance of the behavioral inhibition trait for language development, social interactions, and successful communication, as well as its posited risk for social anxiety disorder, it is important to know whether preschool-age children who stutter are more likely to exhibit behavioral inhibition than their non-stuttering peers and whether this temperamental trait is associated with speech-language characteristics in these children. To the authors' knowledge, only one study has examined BI in preschool-age children who do and do not stutter (Choi, Conture, Walden, Lambert, & Tumanova, 2013). Using direct behavior observation in an interaction with an unfamiliar adult, we found that there was no difference in the degree of BI between the CWS and CWNS. However, when only children in the extreme high and low BI categories were selected, there were significantly more CWS in the high BI group than CWNS. We also reported that CWS in the high BI group stuttered more during a conversation with an unfamiliar adult examiner than CWS in the low BI group. Given the stability of the BI trait and its robust association with several developmental outcomes, including social communication and language development, further study of the potential association between BI and childhood stuttering is warranted.

The present study builds on and extends the Choi et al. (2013) work by recruiting a larger number of preschool-age participants and employing both direct behavioral observation and a

caregiver questionnaire to assess the degree of children's BI. An additional direct behavioral observation measure of BI as well as additional language characteristics were considered in the analyses. Further, to increase precision, acoustic analysis was used to measure durations of all speech segments. Regression models that allowed us to statistically control for the known effects of covariates were employed to test our research questions. Lastly, we also considered additional variables that may be associated with participants' BI as described below.

Purpose of the study. In light of somewhat conflicting evidence from parent-report studies, our goal was to assess behavioral inhibition in preschool-age CWS and CWNS in a social interaction using direct behavior observation procedures established by Kagan and colleagues (Kagan, Reznick, & Snidman, 1987) and used by Choi et al. (2013). Further, the purpose of this study was to examine the associations between the temperamental trait of BI and key language characteristics in all preschool-age children. For CWS, we also assessed the associations between BI and both speech fluency and the negative consequences of stuttering these children may have experienced based on parent-report. Based on the existing research, we hypothesized that the behavioral inhibition trait would be as common in preschool-age CWS as among CWNS. However, we further hypothesized that CWS who were behaviorally inhibited would have less complex language and would speak less with an unfamiliar examiner. As a result, these high BI CWS would stutter less during an interaction with an unfamiliar examiner than those CWS who were not behaviorally inhibited.

The present study sought to answer the following research questions:

1. Do preschool-age children who stutter (CWS) have a higher degree of behavioral inhibition than children who do not stutter (CWNS)?

2. Is there an association between the degree of behavioral inhibition and language output within each group of participants (CWS and CWNS)?
3. Is there an association between the degree of behavioral inhibition and frequency of stuttered disfluencies for CWS?

Method

Sixty-eight preschool-age children (age range: 36–69 months) and their caregivers participated in the study. Participants included 31 CWS (27 boys and 4 girls; mean age 4 years, 1 month) and 37 CWNS (32 boys and 5 girls; mean age 4 years, 3 months). All participants were paid volunteers recruited through an advertisement in a monthly parent magazine circulated throughout Syracuse, New York and an e-mail advertisement sent to Syracuse University employees. The study procedures were approved by the Syracuse University Institutional Review board. Informed consent by parents and verbal assent by children were obtained.

Group classification. Participants were assigned to the CWS group if they (a) produced 3% or more of stuttered disfluencies (i.e., sound/syllable repetitions, sound prolongations, or monosyllabic whole-word repetitions) in a 300 word conversational speech sample (Tumanova, Conture, Lambert, & Walden, 2014; Yaruss, 1997) (b) scored 10 or greater on the SSI-4 (Riley, 2009), and (c) their caregivers expressed concern regarding stuttering. Children whose parents expressed no concern about their child's fluency and who produced less than 3% stuttered disfluencies were assigned to CWNS group. Stuttering frequency and severity characteristics for CWS are presented in Table 1.

Procedures

All data collection procedures took place in the Syracuse University Stuttering Research Laboratory over a single visit. Participants and their caregivers (mothers in the majority of the

cases) were led to the laboratory room, which was unfamiliar to the participants. Upon entry into the laboratory, participants were separated from their caregivers, who stayed in the adjacent room and observed their children over live-stream video. Participants first engaged in a free-play conversation with an unfamiliar female examiner. They were seated at a child-size table beside the examiner and a barnyard toy set was provided to engage the participants in play. The examiner parallel-played with the participant. The conversations were child-directed, but the examiner asked questions or provided prompts when the child was not speaking. Each conversation contained a minimum of 300 words spoken by the participant. After approximately 15 minutes, the free-play conversation ended and participants were administered a standardized test of speech and language (described below). Measures of behavioral inhibition were derived from this free-play conversation speech sample. While the participants engaged in speech-language testing, their caregivers completed the study questionnaires.

Language and hearing abilities. Participants' receptive and expressive language abilities were evaluated using the *Clinical Evaluation of Language Fundamentals – Preschool 2* test (CELF-P2; Wiig, Secord, & Semel, 2005). Language standard scores are presented in Table 2. All participants received a bilateral pure tone hearing screening to rule out hearing impairments with passing levels at 20 dB HL (American Academy of Audiology Task Force, 2011).

Speech sample analysis. Examiner-child interactions were recorded and subsequently transcribed using *Systematic Analysis of Language Transcripts*, instructional version 18 (SALT; Miller & Iglesias, 2017). Standard SALT transcription conventions were followed. Unintelligible speech and stuttered disfluencies were marked in the transcripts according to the SALT conventions. Stuttered disfluency counts were taken only for the first 300 words spoken by the

participants. Sound/syllable repetitions, monosyllabic whole-word repetitions, audible sound prolongations, and blocks (tense pauses) were classified as stuttered disfluencies (Tumanova et al., 2014; Yaruss, 1997).

Behavioral inhibition (BI) assessment. We selected two variables as direct measures of BI, both of which have established reliability and validity (Kagan et al., 1989; Kagan et al., 1999). The first variable was number of spontaneous comments produced. A spontaneous comment was defined as any remark that was not a direct answer to the examiner's question; if a child elaborated an answer, asked the examiner a question, or remarked on an incident in the child's life, a spontaneous comment was coded (Kagan et al., 1989). The duration of the comment was not a factor. Codes for each type of comment were entered in the SALT transcriptions, according to the SALT conventions.

The second variable was the latency to the 6th spontaneous comment, defined as the time from the onset of the child's first utterance of any kind to the onset of the child's 6th spontaneous comment. Onset times were identified on the acoustic waveform using version 2.2.2 of Audacity (R) recording and editing software. Time stamps were measured in seconds with the measurement accuracy to the nearest millisecond. Durations of the participants' stuttered disfluencies and unintelligible utterances (up to the time of the 6th spontaneous comment) were also measured acoustically using Audacity (R). The latency to the child's 6th spontaneous comment was calculated by subtracting the onset time of the child's first utterance of any kind, and the combined durations of the participant's stuttered disfluencies and unintelligible utterances (up to the time of the 6th spontaneous comment) from the onset time of the 6th spontaneous comment, to prevent these from confounding the measure. Consistent with previous research (Choi et al. 2013), the time for the examiner's utterances was included in the time

interval to the 6th spontaneous comment. A longer latency to the 6th spontaneous comment and a smaller number of spontaneous comments were regarded as signs of higher BI (Kagan et al., 1989).

Categorization of BI. Research suggests that BI is best characterized categorically rather than on a continuous dimension (Kagan et al., 1989; Kagan, 1994; Woodward et al., 2000). Thus, consistent with previous research, those participants whose latency to the 6th spontaneous comment were in the top and bottom 15% of the distribution were assigned to high and low BI groups respectively. We chose the fifteenth and eighty-fifth percentiles because previous research (e.g., Kagan et al., 1989; Kagan & Snidman, 2004) revealed that about 15–20% of preschool-age children were consistently inhibited or uninhibited in laboratory assessments. Additionally, using the top and bottom 15% cut-off points allowed us to compare our data to previous research with preschool-age children who stutter, where the same cut-off points were selected (Choi et al., 2013).

Reliability of coding. Two trained coders independently completed SALT transcriptions for 10 randomly selected participants (approximately 15% of the data) and acoustically measured latency to the 6th spontaneous comment for each of the participants. These transcriptions were compared for the MLU measurement, spontaneous comment coding, stuttered disfluency coding, and latency to the 6th spontaneous comment measurement. The reliability of measurement between the two coders was assessed by calculating intra-class correlation coefficients (ICC) using two-way mixed models and absolute agreement criterion (Hallgren, 2012; McGraw & Wong, 1996). The results of these comparisons indicated strong reliability for the first and second coder's MLU measurement (ICC = .981, $p < .0001$), spontaneous comment coding (ICC = .939, $p < .0001$), stuttered disfluencies coding (ICC = .968, $p < .0001$), and the latency to the

6th spontaneous comment (ICC = .988, $p < .0001$). The above ICC reliability values exceed the popular criterion of .7 (Yoder & Symons, 2010).

Parent-report measure of shyness. Participants' temperament was measured with the *Children's Behavior Questionnaire Short Form* (CBQ-SF, Rothbart, Ahadi, Hershey, & Fisher, 2001; Putnam & Rothbart, 2006) which was administered to the caregiver (primarily mothers) who brought the child to the lab. The CBQ-SF was chosen as a parent-report measure of temperament because it has established validity and reliability and has been widely used to assess temperament in preschool-age children (e.g., Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Eggers, De Nil, & Van den Bergh, 2010). The CBQ-SF consists of 94 items scored in the following manner: 1 = Extremely Untrue, 2 = Quite Untrue, 3 = Slightly Untrue, 4 = Neither True or Untrue, 5 = Slightly True, 6 = Quite True, 7 = Extremely True, with a Not Applicable (N/A) option available. The scale rates the child on 15 different behavioral dimensions that combine to form three composite scores known as the CBQ factors: (a) Surgency (activity level, approachability, high intensity pleasure, impulsivity, and shyness), (b) Negative Affectivity (anger/frustration, discomfort, fear, sadness, and soothability), and (c) Effortful Control (attentional focusing, inhibitory control, low intensity pleasure, perceptual sensitivity, smiling and laughter). Although the entire CBQ-SF was administered to the caregivers, we were specifically interested in the CBQ Shyness scale as it reflects a child's tendency to approach novel situations and unfamiliar people. Thus, this scale served as a parent-report measure of BI.

Measure of negative consequences of stuttering. The *Disfluency-Related Consequences* observational rating scale from *the Test of Childhood Stuttering* (TOCS; Gillam, Logan, & Pearson, 2009) was used to assess the negative impact of stuttering on CWS,

consistent with the method of Tumanova et al. (2018). The TOCS *Disfluency-Related Consequences* scale contains nine questions that ask parents to describe the consequences of their child's disruptions in fluency (e.g., questions about what the child does when disfluent or how she/he reacts to speech difficulties in general). Parents are asked to provide their judgements about each behavior (e.g., child shows frustration when disfluent) for their child by rating the frequency of occurrence of these behaviors on a 1–4 ordinal rating scale (i.e., never – rarely – sometimes – often).

Statistical Analyses

The latency to the 6th spontaneous comment, the number of all spontaneous comments, and the CBQ Shyness score served as the dependent variables for research question 1. The mean length of utterances measured in morphemes and number of words spoken during the conversation served as the dependent variables for research question 2. Lastly, the number of stuttered disfluencies during the first 300 words spoken served as the dependent variable for research question 3.

Before conducting the main statistical analyses for each research question, distributions of each dependent variable were visually inspected with histograms and checked for normality based on descriptive statistics (mean, standard deviation, variance, skewness and kurtosis).

Results of the Shapiro-Wilk test of normality indicated that the distributions for the latency to the 6th spontaneous comment ($W = .597$, $df = 37$, $p < .0001$ for CWNS and $W = .814$, $df = 31$, $p < .0001$ for CWS) and number of stuttered disfluencies ($W = .858$, $df = 31$, $p = .001$ for CWS) followed a right-skewed, non-normal distribution. Thus, a univariate generalized linear model with a gamma distribution was fitted to the data to analyze the latency to the 6th spontaneous comment; a univariate generalized linear model with a negative binomial

distribution was fitted to the data to analyze the number of stuttered disfluencies. The number of all spontaneous comments, number of words spoken, and the mean length of utterances followed a normal distribution. Thus, for these dependent variables a univariate general linear model was performed. An alpha level of $p \leq 0.05$ was set for each of the analyses.

Results

Group differences on measures of speech and language are reported first, followed by analyses of each of the research questions.

Age and language skills by group. A multivariate analysis of variance (ANOVA) revealed no significant between-group difference in chronological age ($F_{1,66} = 1.046, p = .310$), or number of words spoken ($F_{1,66} = 1.735, p = .192$). However, CWNS had a higher standard score on CELF-P2 core language ($F_{1,66} = 15.631, p < .0001$) and a higher mean length of utterance ($F_{1,66} = 5.377, p = .024$); as shown in Table 2. Given the variability in the participants' language skill, we controlled for this variable statistically in related analyses.

Research Question 1: Do preschool-age children who stutter have a higher degree of behavioral inhibition than children who do not stutter?

A univariate generalized linear model for the latency to the 6th spontaneous comment as the dependent variable was fitted to the data. CELF-P2 core language score and gender were entered as covariates to account for their possible effects on the dependent variable. The results indicated no significant difference in the degree of BI between preschool CWS and CWNS (Wald $\chi^2 = .048, df = 1, p = .826, d = .231, \beta = 5.824$ for CWNS). There was a significant effect of CELF-P2 core language score (Wald $\chi^2 = 11.212, df = 1, p = .001, \beta = -2.876$) on the latency to the 6th spontaneous comment, such that participants who had lower CELF-P2 core language scores took longer to produce the 6th spontaneous comment. Although the effect of gender on the

latency to the 6th spontaneous comment was not significant (Wald $\chi^2 = 3.041$, $df = 1$, $p = .081$), a standardized regression coefficient beta ($\beta = 93.438$) indicated a trend of girls taking longer to produce their 6th spontaneous comment than boys.

Similarly, the results of the univariate general linear model revealed no significant difference in the number of spontaneous comments produced by CWS and CWNS ($F_{1,64} = .003$, $p = .954$, $\eta^2_p < .0001$). Gender had a significant effect on the number of spontaneous comments ($F_{1,64} = 3.903$, $p = .053$, $\eta^2_p < .057$). A standardized regression coefficient beta ($\beta = -30.825$) indicated that girls produced fewer spontaneous comments than boys. The effect of the CELF-P2 core language score on the number of spontaneous comments was not significant ($F_{1,64} = .178$, $p = .674$, $\eta^2_p = .003$). Table 2 presents descriptive data for the dependent variables.

Cross tabulation analysis of the high and low BI groups (top and bottom 15% of the latency to the 6th spontaneous comment distribution) indicated that there was no significant difference in the number of CWS vs. CWNS classified as high or low BI (Pearson $\chi^2 = 1.818$, $df = 1$, $p = .178$, Cramer's $V = .302$); see Table 3.

However, for parent-report measure of BI as the dependent variable, a univariate ANOVA revealed a significant difference in CBQ Shyness score between CWS and CWNS ($F_{1,66} = 5.936$, $p = .018$, $\eta^2_p = .083$, $\beta = -.749$), with parents of CWS rating their children higher on the Shyness scale than did parents of CWNS. The effect size for the group difference in CBQ Shyness score, Cohen's $d = .593$, was medium.

Research Question 2: Is there an association between the degree of behavioral inhibition and language output within each group of participants (CWS and CWNS)?

A univariate general linear model with the mean length of utterances as the dependent variable and the latency to the 6th spontaneous comment, CELF-P2 core language score, and

TOCS *Disfluency-Related Consequences* scale score as covariates was fitted for CWS. For CWNS, the model only included the latency to the 6th spontaneous comment and the CELF-P2 core language score. For CWS, there was significant main effect of the latency to the 6th spontaneous comment on the mean length of utterances ($F_{1,26} = 7.281, p = .012, \eta^2_p = .219, \beta = -.003$; see Figure 1). CWS who had a longer latency to the 6th spontaneous comment (higher BI) demonstrated lower mean length of utterances during the conversation with the unfamiliar examiner. The effects of the CELF-P2 core language score ($F_{1,26} = 1.490, p = .233, \eta^2_p = .054, \beta = .017$; see Figure 2) and the TOCS *Disfluency-Related Consequences* scale score ($F_{1,26} = 1.874, p = .183, \eta^2_p = .067, \beta = -.045$) were not significant. For CWNS, the model indicated no significant main effects of either the latency to the 6th spontaneous comment ($F_{1,34} = .003, p = .955, \eta^2_p < .001, \beta < .0001$; see Figure 1), or the CELF-P2 core language score ($F_{1,34} = 2.359, p = .134, \eta^2_p = .065, \beta = .027$; see Figure 2) on the mean length of utterances produced during the conversation.

A univariate general linear model with the number of words spoken as the dependent variable and the latency to the 6th spontaneous comment and the CELF-P2 core language score as covariates was fitted for each of the groups (CWS and CWNS). For CWS, the model indicated a significant main effect of the latency to the 6th spontaneous comment ($F_{1,28} = 11.370, p = .002, \eta^2_p = .289, \beta = -.685$), such that CWS who had a longer latency to the 6th spontaneous comment (higher BI) said fewer words during the conversation with the unfamiliar examiner. The effect of the CELF-P2 core language score on the number of words spoken was not significant ($F_{1,28} = .020, p = .890, \eta^2_p = .001, \beta = -.385$). For CWNS, the model did not indicate significant main effects of either the latency to the 6th spontaneous comment ($F_{1,34} = 2.287, p = .140, \eta^2_p = .063, \beta$

= -.440), or the CELF-P2 core language score ($F_{1,34} = 1.287, p = .265, \eta^2_p = .036, \beta = 4.582$) on the number of words spoken.

Research Question 3: Is there an association between the degree of behavioral inhibition and frequency of stuttered disfluencies for CWS?

A univariate generalized linear model was fitted to the data. The number of stuttered disfluencies per first 300 words spoken was the dependent variable; the latency to the 6th spontaneous comment and MLU during the conversation with an unfamiliar examiner were entered as covariates. The model indicated a significant main effect of the latency to the 6th spontaneous comment (Wald $\chi^2 = 5.716, df = 1, p = .017, \beta = -.002$) and MLU (Wald $\chi^2 = 4.838, df = 1, p = .028, \beta = -.244$) on the number of stuttered disfluencies produced, such that CWS who had a longer latency to the 6th spontaneous comment (higher BI) had shorter and less complex utterances and produced fewer stuttered disfluencies compared to CWS who showed shorter latencies to the 6th spontaneous comment (lower BI).

Ancillary Analyses

The association between two behavioral measures of BI. To establish agreement with the existing research (Kagan, Reznick, & Gibbons, 1989), we examined the association between the two direct measures of BI employed in our study. Correlational analysis for the two variables used to index BI, namely the latency to the 6th spontaneous comment and the number of all spontaneous comments was significant (Pearson $r = -.403, p = .001$), which is consistent with data reported by Kagan et al. (1989).

The association between behavioral and parent-report measures of BI. Direct behavior observation in a laboratory-based assessment and parent-report of child's behavior have both been used to identify BI in preschool-age children. The latter method, however, is less time

consuming and, thus, has a clinical advantage. The clinical utility of parent report as a means to assess BI is also supported by the strong correlations between this method and direct behavior observation of BI (Bishop, Spence, & McDonald, 2003; Kagan, Reznick, Clarke, Snidman, & Garcia Coll, 1984). The above considerations prompted us to examine whether parent report of BI (CBQ Shyness score) and the direct behavior observation of BI (the latency to the 6th spontaneous comment) were significantly correlated in our dataset. We chose to examine the association between these two variables separately for each group (CWS and CWNS). This was done to account for the possibility that parents of CWS may be influenced in their responses by their child's stuttering.

Correlational analysis indicated a significant association between the latency to the 6th spontaneous comment and parent-report CBQ Shyness score in CWS (Pearson $r = .561$, $p = .001$) and in CWNS (Pearson $r = .365$, $p = .013$). These results indicate that, for both CWS and CWNS, there is an agreement between objective behavioral measures of BI and subjective caregiver ratings. However, the agreement between the objective behavioral measure and subjective caregiver rating of BI was stronger for CWS, indicating that ratings given by parents of CWS were more consistent with the objective behavioral measures than ratings of parents of CWNS.

Discussion

Overall, the results partially supported our hypotheses related to BI in CWS. Behavioral inhibition was associated with lower mean length of utterances and fewer words spoken for CWS only, meaning that similar links were not found for CWNS. Interestingly, for CWS, BI was also associated with less complex utterances and fewer stuttered disfluencies produced. Finally, there was also evidence that CWS had a higher degree of behavioral inhibition than CWNS, although

this finding was restricted to parent-reported BI and did not extend to observational measures of this characteristic.

The current study advanced our understanding of BI as it relates to childhood stuttering. Specifically, we found that for CWS, higher BI was associated with less complex utterances and fewer words spoken during the conversation with an unfamiliar examiner. This finding adds to a growing number of studies that report associations between the BI trait and language characteristics in preschool-age children (Harrison & McLeod, 2010; Paul & Kellogg, 1997). For example, Paul and Kellogg (1997) found that more outgoing first grade children tended to show longer mean length of utterances than their less outgoing peers. Further, first grade children who had a history of slow expressive language development have been consistently rated as more shy and withdrawn, leading the authors to suggest that this temperamental trait contributed to their slow language development (Paul & Kellogg, 1997). Similar results were obtained in a population-wide study of 4- and 5-year old Australian children (Harrison & McLeod, 2010). Their findings indicated that “temperamental sociability” (consistent with low BI) was associated with a reduced risk of speech and language impairment, whereas “higher reactivity” and “low approach” (consistent with high BI) were associated with greater risk of speech and language impairment. More recently, similar findings have been reported in younger preschool-age children (24- to 30-month old in age) by Usai, Garello and Viterbori (2012).

Despite the significant association between the degree of BI and language characteristics of CWS, there was no association between degree of BI and language outcomes in the CWNS. Based on a visual inspection of the data, we can speculate that the lack of the association between BI and language output in CWNS as compared to CWS may be due to a lower variability in the latency to the 6th spontaneous comment observed in CWNS.

A body of research indicating BI trait's regulatory influence on young children's behavior (including language development and social adaptation), taken together with our findings of the association between the degree of BI and CWS's language characteristics, suggest that this trait may be clinically relevant in several ways. Those preschool-age children who stutter and are behaviorally inhibited may be less able to cope with difficulties in social interactions resulting from stuttering. For these children, the behavioral inhibition trait could amplify the negative consequences of stuttering; it could affect not only these children's social relationships, but also their language and social-emotional development. Given the possibility of these undesirable consequences of the BI trait, we suggest that clinicians working with preschool-age children who stutter consider gathering information about children's degree of BI during the assessment for stuttering. Both direct behavior observation and parent-report questionnaires have been used to assess children's degree of BI, however, the latter method has several clinical advantages. Parent-report questionnaires are typically easy to administer and score, and require minimal time commitment from the clinician. In contrast, direct behavior observation assessment, such as the one employed in this study, is time-consuming and costly. Given these considerations, published caregiver-report questionnaires with established psychometric properties (e.g., Bishop et al., 2003) may be a more feasible screening method that could be used to identify children's degree of BI in a clinical setting.

Related to the above discussion of the role of BI in childhood stuttering, we found that there was a significant association between the degree of BI and the frequency of stuttered disfluencies produced by CWS during a conversation with an unfamiliar adult examiner. Contrary to some of the findings reported by Choi et al. (2013), we found that preschool-age CWS with higher BI produced fewer stuttered disfluencies during the first 300 words of the

conversation with an unfamiliar examiner than CWS with lower BI. The known effect of linguistic complexity on stuttering frequency (Bernstein Ratner & Sih, 1987; Logan & Conture, 1995, 1997; Sawyer et al., 2008; Zackheim & Conture, 2003) could potentially explain the differences in our and Choi et al.'s findings. The Choi et al.'s analytical procedure did not allow for language complexity to be considered in the analysis. In the present study, CWS who had a higher BI produced shorter MLUs and fewer words and hence demonstrated fewer stuttered disfluencies than CWS who were low BI. Considering the Salley et al. (2013) report that higher levels of shyness in situations involving novelty were strongly associated with impairments in both social motivation and social communication in 3 and 4 year-old children, our finding suggests that CWS who have a higher degree of BI may curtail their talking in a novel situation with an unfamiliar examiner. Indeed, withdrawal from talking was the most frequently reported reaction to stuttering in preschool-age children in a sample of 1,122 parents of preschool-age children who stutter (Boey et al., 2009).

The findings that CWS who had a higher BI produced shorter MLUs, fewer words, and fewer stuttered disfluencies than CWS who were low BI have important implications for diagnosing stuttering in young children. As CWS are more likely to stutter when producing syntactically complex utterances than syntactically simple utterances (e.g., Bernstein Ratner & Sih, 1987; Logan & Conture, 1995, 1997; Sawyer et al., 2008; Zackheim & Conture, 2003), saying less in a diagnostic evaluation may lead to fewer moments of stuttering and potentially an incorrect diagnosis of typical fluency. In other words, given that fear of novelty and inhibition have been shown to inhibit social relationships (Salley & Dixon, 2007), those CWS who are shy may talk less during a diagnostic stuttering evaluation, masking their true stuttering severity. During a diagnostic evaluation for these children, it may be necessary to collect several speech

samples, including some speech samples with familiar people and in familiar surroundings (such as talking with a caregiver at home or at school).

Having discussed the implications of high behavioral inhibition on speech and language characteristics of CWS, we would like to focus on our findings related to whether BI trait is more common in preschool-age CWS. Our direct behavior observation measures indicated that preschool-age CWS are as likely to have a temperamental trait of behavioral inhibition as preschool-age CWNS. The mean latency to the 6th spontaneous comment was longer for CWS, but this difference did not reach significance. Similarly, there was no significant difference in the number of spontaneous comments produced by CWS and CWNS. Additionally, the number of CWS and CWNS in the high vs. low BI categories was not significantly different. Despite the lack of significant differences between CWS and CWNS in these direct measures of BI, parents rated CWS significantly higher on the CBQ Shyness scale, which reflects a higher degree of BI. These somewhat conflicting results are consistent with other reports in the literature. Specifically, Choi et al. (2013) found no difference in the direct behavioral measure of BI, namely the latency to the 6th spontaneous comment, between preschool-age CWS and CWNS in their study.

The higher rating of shyness by parents of CWS does not necessarily contradict our direct behavior observation results. Given some concerns about the accuracy of parent-report of a child's behavior (e.g., Kagan, Snidman, McManis, Woodward, & Hardway, 2002), we acknowledge the possibility that the parents of CWS in the present study may have described their children's temperament differently because of their child's stuttering. However, there is ample research evidence of agreement between direct behavior observation and parent-report of BI (Bishop, Spence, & McDonald, 2003; Henderson, & Wachs, 2007; Kagan, Reznick, Clarke,

Snidman, & Garcia Coll, 1984). Our data also provide evidence of agreement between direct behavior observation and parent-report of BI, especially for CWS, as the two measures were moderately correlated in our sample.

Moreover, it is important to acknowledge that the BI construct has situational specificity, meaning that children who have a high BI can display an avoidant behavior in several different contexts but not necessarily in all of them (Kagan et al., 1998; Bishop et al., 2003). Parents have the potential for providing a more general tendency of their child's behaviors and capturing the situational specificity in the expression of BI, as they routinely observe their child across a variety of changing circumstances. We, on the other hand, observed our participants in a single situation, namely the interaction with an unfamiliar adult examiner. This leaves the possibility, for example, that the participants can show a higher degree of BI when they interact with peers as opposed to adults or in a group of people as opposed to in one-on-one interaction. Our parent-report results of higher shyness in preschool-age CWS are also in line with the results of two recent large-scale studies of preschool-age CWS temperament (Briley et al., 2019; McAllister, 2016). Together, these results highlight the need for practitioners to carefully consider children's temperament, and their degree of shyness specifically, in clinical diagnosis and treatment for stuttering, as others have already suggested (Conture, Kelly, Walden, 2013; Jones, Choi, Conture, & Walden, 2014). In the paragraphs that follow, we offer some suggestions for how to consider CWS's degree of BI in the treatment for stuttering.

Developmental and clinical research in child psychology offers insight on treatment considerations for young children who stutter and have high BI. In a recent review, Chronis-Tuscano, Danko, Rubin, Coplan and Novick (2018) discussed the role of parenting style as an important and modifiable factor that can decrease the risk of development of social anxiety

disorders in young children who have high BI. Specifically, multiple studies reviewed by Chronis-Tuscano et al. (2018) found that an overprotective parenting style was associated with an increased risk of the development of social anxiety disorders in children who have high BI. A less restrictive, autonomy-granting parenting style, on the other hand, acted as a protective factor. Regarding the treatment considerations, Chronis-Tuscano et al.'s (2018) review indicated that parenting interventions emphasizing gentle encouragement of approach behaviors and cognitive-behavior therapy for children, as young as preschool-age, and their parents may prevent social anxiety development in children who have high BI. Notably, cognitive-behavior therapy has also been found to be an effective treatment for stuttering in children, adolescents, and adults (e.g., Beilby & Byrnes, 2012; Kelman & Wheeler, 2015; Menzies, Onslow, Packman, & O'Brian, 2009; Nnamani et al., 2019). Cognitive-behavior therapy for preschool-age children has to be adapted and play-based to suit the young age of the client (Hirshfeld-Becker et al., 2008; 2010; Knell, 2015). Given the effectiveness of this intervention in preventing anxiety in the at-risk preschool-age population, clinicians are encouraged to consider incorporating cognitive-behavior therapy when working with CWS who have high BI as well as including caregiver counseling in the treatment program for these children (Rapee, Kennedy, Ingram, Edwards, & Sweeney, 2005).

Taken together, the pattern of results reported here may have important clinical implications. Specifically, those CWS who have a higher degree of BI may speak less, and thus stutter less, during a diagnostic evaluation for stuttering, making it harder for the clinician to establish the correct diagnosis for stuttering. These same children, however, may be at a greater risk of negative impact of stuttering on their social communication and social development.

Present results support continued study and further consideration of the association between CWS's temperament, social development, and stuttering development trajectories.

Future Directions

Young CWS may experience negative peer responses to their speech beginning in the preschool years (Langevin, Packman, & Onslow, 2010). Difficulties with interpersonal communication put these children at an increased risk of negative social and psychological impacts (Iverach et al., 2011). As shyness is associated with socioemotional maladjustment even at the preschool age (Coplan & Armer, 2005), those children who stutter and are behaviorally inhibited may be at a greater risk for the negative impact of stuttering. When faced with negative experiences during social interactions with peers, CWS who are shy and highly reactive to novelty, compared to those who are low BI, may experience a stronger adverse impact of stuttering leading to increased social withdrawal and difficulties with socialization and coping. As low BI may be a protective factor for social and language development, it may also serve as a protective factor for stuttering development. Conversely, high BI may serve as a risk factor. Future research is required to test these hypotheses.

Conclusion

This study provides empirical evidence that behavioral inhibition to the unfamiliar (also known as shyness) may have salience for childhood stuttering, as it affected both the quantity and quality of preschool-age CWS's language spoken with an unfamiliar adult. The present findings are consistent with the notion that during the assessment of stuttering, clinicians should also evaluate preschool-age children's temperament, particularly their degree of shyness or behavioral inhibition to novelty. Published caregiver-report questionnaires with established psychometric properties offer a feasible method to screen for children's degree of behavioral

inhibition in a clinical setting. Further, as the behavioral inhibition trait could have an adverse effect on children's language and social-emotional development and could amplify the negative consequences of stuttering, we suggest that clinicians consider including caregiver counseling and elements of cognitive-behavior therapy in the treatment program for preschool-age CWS who have high behavioral inhibition.

Known for its situational variability, stuttering is most apparent in social contexts, especially in interactions with unfamiliar people. Given that behavioral inhibition is a stable trait that affects how people approach novel and social situations, further investigation on the role of the behavioral inhibition trait on stuttering is warranted.

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Table 1: Stuttering severity, assessed by the Stuttering Severity Instrument – 4 (SSI-4; Riley, 2009) and the Test of Childhood Stuttering Disfluency-Related Consequences Rating Scale (TOCS DRC; Gillam, Logan, & Pearson, 2009) for children who stutter (CWS).

Participant Number	Group	Gender	TOCS DRC score	Stuttering Frequency (%)	SSI-4 score	Stuttering severity
1	CWS	F	12	9	21	moderate
2	CWS	F	8	7	15	mild–moderate
3	CWS	F	2	3	14	mild–moderate
4	CWS	F	7	3	16	mild–moderate
5	CWS	M	13	7	20	moderate
6	CWS	M	13	8	16	mild–moderate
7	CWS	M	4	9	16	mild–moderate
8	CWS	M	0	7	14	mild–moderate
9	CWS	M	12	5	18	moderate
10	CWS	M	5	8	16	mild–moderate
11	CWS	M	18	7	16	mild–moderate
12	CWS	M	5	4	12	mild
13	CWS	M	9	22	29	severe
14	CWS	M	7	9	18	moderate
15	CWS	M	0	5	12	mild
16	CWS	M	0	8	18	moderate
17	CWS	M	12	6	15	mild–moderate
18	CWS	M	9	14	20	moderate
19	CWS	M	0	8	20	moderate
20	CWS	M	3	12	22	moderate
21	CWS	M	4	7	18	moderate

22	CWS	M	5	4	10	very mild—mild
23	CWS	M	8	4	12	mild
24	CWS	M	6	9	16	mild—moderate
25	CWS	M	5	4	10	very mild—mild
26	CWS	M	8	14	20	moderate
27	CWS	M	5	5	14	mild—moderate
28	CWS	M	6	4	10	very mild—mild
29	CWS	M	7	3	10	very mild—mild
30	CWS	M	2	4	14	mild—moderate
31	CWS	M	5	4	10	very mild—mild

Table 2: Descriptive statistics for age, measures of language, and BI for children with do (CWS, n = 31) and do not stutter (CWNS, n = 37).

Independent variable	Group	Mean	Std. Deviation	Difference Significant
Age (months)	CWNS	50.89	9.35	<i>n.s.</i>
	CWS	48.61	8.91	
CELF-P2 Core Language Standard Score	CWNS	112.92	10.05	$p < .0001$
	CWS	103.16	10.24	
Mean Length of Utterances during conversation	CWNS	4.15	1.03	$p = .024$
	CWS	3.6	.88	
Number of Words Spoken during conversation	CWNS	580.54	35.62	<i>n.s.</i>
	CWS	511.03	38.92	
Latency to the 6 th spontaneous comment (seconds)	CWNS	114.04	139.64	<i>n.s.</i>
	CWS	146.18	138.70	
Number of spontaneous comments	CWNS	87.59	40.11	<i>n.s.</i>
	CWS	89.29	47.85	
CBQ Shyness Score	CWNS	3.45	1.24	$p = .018$
	CWS	4.19	1.26	

Note: CELF P2 = Clinical Evaluation of Language Fundamentals Preschool Version 2 Test; *n.s.* = not statistically significant.

Table 3: Cross Tabulation analysis for CWS and CWNS in the high vs. low BI categories, N = 20.

High/Low BI group	Low BI (bottom 15%)	High BI (top 15%)	Chi Square	Difference Significant
CWNS	6 (4.5)	3 (4.5)	1.818	<i>n.s.</i>
CWS	4 (5.5)	7 (5.5)		

Note: Expected values are enclosed in parentheses below the observed values.

Figure 1: Association between the mean length of utterance (MLU) during conversation and the latency to the 6th spontaneous comment for children who stutter (Group 1; n = 31) and children who do not stutter (Group 0; n = 37).

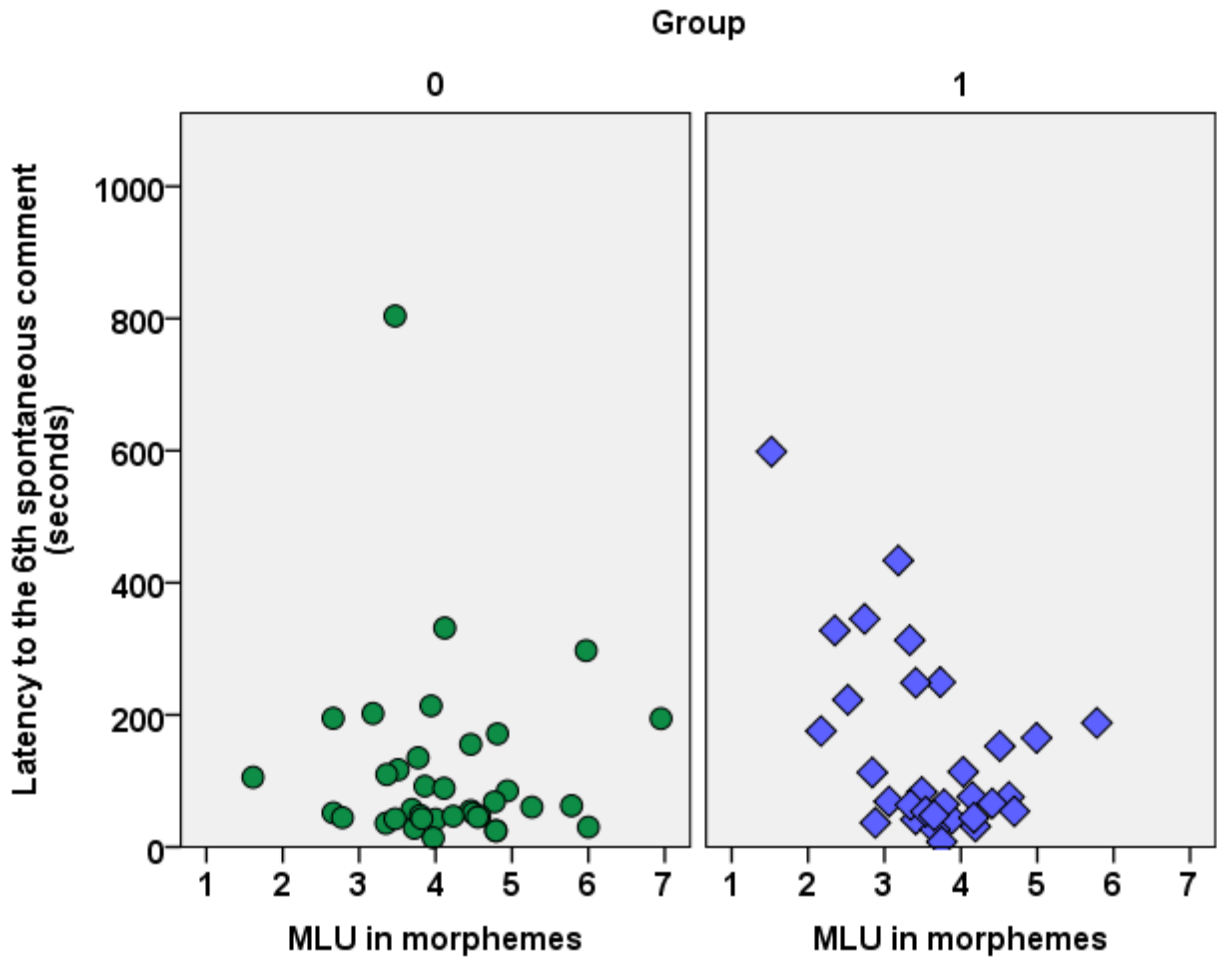


Figure 2: Association between the mean length of utterance (MLU) during conversation and the CELF-P2 Core Language standard score for children who stutter (Group 1; n = 31) and children who do not stutter (Group 0; n = 37).

