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Autonomic Nervous System Response to Speech Production in Stuttering and Normally Fluent Preschool-Age Children

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Abstract

Purpose: We studied speech-related sympathetic nervous system arousal of preschool-age children who do (CWS) and do not stutter (CWNS) and its association with children's proclivity to experience negative emotions, and children's self-reported attitudes towards speaking.

Method: Electrodermal activity measures were collected from 32 preschool-age children while they engaged in a picture description and a non-word repetition task. Children's proclivity to experience negative emotions was assessed with a parent-report questionnaire. Children's communication attitude was assessed with a self-report questionnaire.

Results: CWS did not differ from CWNS in their sympathetic arousal during a picture description task. However, during a more challenging non-word repetition task, preschool-age CWS had a higher sympathetic arousal level than CWNS. Although CWS were rated by their caregivers as more fearful and prone to sadness, children's tendency to experience stronger and more frequent negative emotions was not associated with their sympathetic arousal during speaking. Lastly, although CWS had a more negative communication attitude than CWNS, it was not associated with their level of sympathetic arousal during speaking.

Conclusions: Our findings suggest that age-appropriate social communication tasks are not inherently more stressful for preschool-age CWS and are not associated with state-related stress or anxiety that is often reported for adults who stutter. However, speaking tasks that place a higher demand on children's cognitive-linguistic system may be more taxing and challenging to preschool CWS, leading to a higher level of arousal.

The association between emotional processes and stuttering has been a focus of prior stuttering research (e.g., Brutten & Shoemaker, 1967; Conture & Walden, 2012; Craig, Hancock, Tran, & Craig, 2003; Eggers, De Nil, & Van den Bergh, 2010; Sheehan, 1958), but the role of emotional processes in the etiology and development of stuttering in children is still not well understood. Research into emotional processes has traditionally distinguished between "state" and "trait" emotions. The former are defined as variable, contextually-determined emotional processes related to various situational challenges, whereas the latter are often referred to as temperament and defined as relatively stable, biologically-based individual differences in reactivity and self-regulation (Derryberry & Rothbart, 1984). Presently, not much is known about how either “trait” (temperament) or “state” (contextually-determined) emotional arousal affect speaking and stuttering, however, it is commonly believed that people who stutter hold negative communication attitudes and often associate speaking with negative emotions such as anxiety. Prior research investigating emotional processes and stuttering in children and adults have relied heavily on self-report and parent-report measures (in the case of children who stutter) to assess emotional processes associated with stuttering. Psychophysiological measures offer an objective way to assess “state” emotions, such as an emotional arousal during a specific speaking task. These measures are also complementary to parent-report measures of “trait” emotions for preschool-age children, whose young age precludes them from describing their personality and emotional states reliably. Thus, the purpose of the present study was to investigate potential differences in physiological reactivity of preschool-age children who do and do not stutter during speaking tasks and examine whether the physiological reactivity of these children is associated with their temperamental traits and attitude towards speaking. What follows is an overview of the autonomic nervous system activity in response to emotionally salient stimuli, means to measure this activity and the potential role of physiological reactivity in speaking and the development of stuttering. Further, we consider two psychological constructs, namely temperament and communication attitude, and their potential influence on physiological reactivity in speaking. We end the introduction with our research questions and hypotheses.

Neurophysiological research suggests that all emotions can be described as a combination of two neurophysiological dimensions, valence and arousal (e.g., Russel, 1980; Posner et al., 2009). The valence system determines the degree to which an emotion is pleasant or unpleasant, and the arousal system determines the degree to which a given stimulus is behaviorally activating (e.g., ranging from bored, relaxed, or calm to excited, anxious, or stressed). Neurophysiologic research indicates that activity of the autonomic nervous system is particularly relevant to emotional arousal, cognitive effort or stress. Specifically, whereas the activity of the autonomic nervous system is not sensitive to the valence of the stimulus (e.g., pleasant or unpleasant), it is sensitive to the level of arousal elicited by a given stimulus. Thus, measures of the activity of the autonomic nervous system are useful to objectively assess the degree of speaker's emotional arousal or cognitive effort that is elicited by a certain task (such as speech production). Autonomic nervous system measures may also capture processes that are covert or nonconscious, thus they are especially beneficial for the study of preschool-age children's speaking related reactions.

Autonomic nervous system activity. The sympathetic branch of the autonomic nervous system prepares the body for action in response to environmental stimuli and is known to activate during times of stress (Boucsein, 2012; Dawson, Schell, & Filion, 2000; El-Sheikh et al., 2009). Sympathetic activation increases heart rate, dilates bronchioles, and redistributes blood flow to the muscles. Sympathetic activation also increases the activity of the eccrine sweat glands, which are innervated solely by the sympathetic nervous system (Fowles, 1993). Eccrine sweat gland activity at the skin leads to sweat secretion and a subsequent increase in the skin's electrical conductance. Thus, sympathetic nervous system activity can be reliably indexed by measuring the degree of electrodermal activity. The electrodermal activity measurements are traditionally divided into two types of activity: tonic, such as skin conductance level (SCL), and phasic, such as frequency of skin conductance responses (SCR). Tonic measurements are obtained during intervals when participants are not presented with any specific stimuli designed to elicit a response (Bouscein, 2012). These measurements also include "nonspecific" phasic changes in electrodermal activity that occur spontaneously. Phasic responses, in contrast, are obtained when participants exhibit fluctuations in their electrodermal activity that are linked to specific stimuli that were presented. These phasic responses are time-locked to the presentation of the specific stimuli. Both tonic and phasic measures of electrodermal activity are often employed concurrently to estimate the sympathetic nervous system activity, an important component of a body's response to a situational challenge. Both SCL and SCRs are reliable and valid indices of sympathetic nervous system activity (Bouscein, 2012), and given their non-invasive nature they have been widely used in studies with children as reviewed in the following paragraphs (e.g. El-Sheikh, 2007; Fowles, Kochanska, & Murray, 2000; Nikolić, Aktar, Bögels, Colonnesi, & de Vente, 2018).

Speech production requires complex coordination of movements between respiratory, phonatory and articulatory structures and simultaneous processing of cognitive-linguistic information. Additionally, communicative speech production, such as a conversation with an unfamiliar adult, may present a social communicative challenge for a young child. Indeed, this task has often been used in psychophysiological and personality research with young children to evoke social challenge (e.g. Nikolić, Aktar, Bögels, Colonnesi, & de Vente, 2018). Research shows that speech production leads to increased autonomic arousal in both adults (Het, Rohleder, Schoofs, Kirschbaum, & Wolf, 2009; Kirschbaum, Pirke, & Hellhammer, 1993; Weber & Smith, 1990), and children (Arnold, MacPherson, & Smith, 2014; Kleinow & Smith, 2006). Moreover, autonomic arousal levels for speech exceed those of high-effort non-speech tasks such as the Valsava maneuver (Weber & Smith, 1990) or taking a test of intelligence (Peters & Hulstijn, 1984). Given that children's speech-motor and linguistic abilities are still developing compared to those of adults (especially those of younger, preschool-age, children), speaking may present a greater challenge for them than for adults. Although no studies directly compared the level of sympathetic arousal during speech production between preschool-age children and adults, Kleinow and Smith (2006) reported that school-age children demonstrated higher level of sympathetic arousal (as indexed by higher SCL) during a sentence repetition task than adults.

Considering the evidence that children who stutter (CWS) may lag in their speech motor control skill development (Smith, Goffman, Sasisekaran, & Weber-Fox, 2012; MacPherson & Smith, 2013) and have some areas of lower language performance than their non-stuttering peers (Ntourou, Conture, & Lipsey, 2011), speech production may be more challenging for them and result in elevated levels of autonomic arousal compared to children who do not stutter (CWNS). There have only been a few published studies of autonomic arousal of preschool-age CWS and CWNS during speech production, all conducted by the same research lab (Choi et al., 2016; Jones et al. 2014; Zengin-Bolatkale, Conture & Walden, 2015, 2018). Jones et al. (2014) measured respiratory sinus arrhythmia (an indicator of parasympathetic autonomic nervous system activity) and skin conductance level in preschool-age children while they watched positively- and negatively-valenced video clips and during picture description tasks immediately after video viewing. They reported that CWS, compared to CWNS, only demonstrated a higher SCL during picture description tasks subsequent to viewing of a positively-valenced video clip, but not subsequent to viewing of negative or neutral video clips (neutral video clips were used to establish the baseline for autonomic nervous system measures). Zengin-Bolatkale, Conture and Walden (2015) measured SCL in preschool-age children during a time-pressure picture-naming task, where the children were asked to name pictures of common objects as fast as possible. There was no between-group difference in SCL during the picture-naming task overall, however some differences emerged when participants were divided into specific age groups (e.g., 3, 4 and 5 year-olds). Three-year-old CWS demonstrated a significantly higher SCL than 3-year-old CWNS, whereas there were no differences in SCL for 4- and 5-year-olds. These mixed findings are somewhat difficult to interpret and warrant more research into the autonomic arousal of preschool-age CWS and CWNS during speech production. Additionally, research suggests that such factors as temperament and communication attitude may contribute to the autonomic arousal of preschool-age children. These factors and their significance for preschool-age children who stutter are reviewed below.

Temperament. A child's temperament may affect their level of autonomic arousal during speech. Temperament is defined as relatively stable, biologically-based individual difference in reactivity and self-regulation (Derryberry & Rothbart, 1984). Children who have a proclivity for emotional reactivity may be more susceptible to contextually-determined challenges. Several studies found relations between temperamental qualities and sympathetic nervous system activity in children (Fowles, Kochanska, & Murray, 2000; Kagan 1997, Nikolić, de Vente, Colonnesi, & Bögels, 2016; Nikolić, Aktar, Bögels, Colonnesi, & de Vente, 2018). For example, Nikolić et al. 2018 reported that preschool-age children's level of arousal (measured by electrodermal activity and heart rate variability) during a conversation with a stranger when they were 4.5 years of age was associated with their later reports of social anxiety assessed when they were 7 years of age. Electrodermal activity is one of the primary measures that has been used to relate temperament and sympathetic responsivity, and some recommend this physiologic variable instead of questionnaire data as a main indicator of reactivity to medium intensity stressors (Katkin, 1975).

The role of temperament in stuttering development has received considerable attention in recent years. Although there is no clear evidence that temperament plays a causal role in stuttering (Alm, 2014; Kefalianos, Onslow, Block, Menzies, & Reilly, 2012), some converging research findings based on caregiver reports and direct behavior observation point to differences in temperament between preschool-age CWS and CWNS. CWS have been reported to exhibit lower attentional control (e.g., Eggers, De Nil, & Van den Bergh, 2010, 2012, 2013; Schwenk, Conture, & Walden, 2007), higher emotional reactivity (Anderson, Pellowski, Conture & Kelly, 2003; Choi, Conture, Walden, Jones, & Kim, 2016; Karrass et al., 2006) and greater negative affect than CWNS based on parent-report (Ambrose, Yairi, Loucks, & Seery, 2015; Eggers, De Nil, & Van den Bergh, 2010) and direct behavior observation (Johnson et al., 2010; Ntourou, Conture, & Walden, 2013). Notably, using the Children’s Behavior Questionnaire Short Form (CBQ; Putnam & Rothbart, 2006) to assess preschool-age children’s temperament, Ambrose, Yairi, Loucks, and Seery (2015) reported that children who did not recover from stuttering by the 4-year follow-up after the original diagnosis had significantly higher scores on the temperamental construct of Negative Affectivity than those children who recovered from stuttering and those children who never stuttered.

Young children's temperamental qualities related to "Negative Affectivity" (Derryberry & Rothbart, 1984, pp.132-166) have previously been identified as a risk factor for the development of anxiety disorder (Cote et al., 2009). Cote et al. (2009, pp.1204) defined children who are at risk as often displaying the following attributes: "nervous, high strung or tense", "appears fearful or anxious", "appears worried", "not as happy as other children", and "has difficulty having fun." In light of Cote et al. (2009) and Ambrose et al. (2015) findings, it is important to know whether preschool-age CWS are more likely to exhibit a higher degree of Negative Affectivity than their normally fluent peers, as it can put them at risk of developing an anxiety disorder at a later time. As reviewed in the paragraphs below, anxiety may be a result of stuttering disorder itself, but the directionality of the proposed association is not yet clear. Studying these temperamental qualities in very young children who stutter has the potential to elucidate the proposed association. It also remains unclear if preschool-age children's proclivity for experiencing stronger and more frequent negative emotions is associated with higher sympathetic nervous system activity during novel, potentially stressful speaking situations.

Negative communication attitude. Adolescents and adults who stutter frequently report subjective feelings of anxiety towards social communication. They tend to perceive themselves as incompetent communicators, finding communication difficult and feeling apprehensive about talking, which may be interpreted as signs of a negative cognitive bias towards communication. Defined as a tendency to preferentially process negatively valenced information, negative cognitive bias has been considered by many to play a central role in the onset and maintenance of anxiety (Beck, 2008; Mathews & MacLeod, 2005; Rapee & Heimberg, 1997; Wong & Rapee, 2016). Multiple research studies demonstrate that adults and adolescents who stutter frequently report speaking-related anxiety (Craig & Tran, 2014; Gunn et al., 2014; Iverach & Rapee, 2014;

Messenger, Onslow, Packman, & Menzies, 2004; Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014), and can show clinical signs of a social anxiety disorder (Blumgart, Tran, & Craig, 2010; Iverach, Jones, et al., 2016; Iverach, O'Brian, et al., 2009; Menzies et al., 2008; Stein, Baird, & Walker, 1996). For adults and adolescents who stutter, however, it is difficult to distinguish the anxiety that results from the stuttering disorder from the anxiety that may have been driven by personality-related factors. Examining these processes in young, preschool-age, children may elucidate the origins of speech-related anxiety in this population.

Although it is not clear when children who stutter start associating speech production with negative emotions such as anxiety or stress, research suggests that these emotions develop as a consequence of stuttering, presumably due to an increased risk of negative social and psychological impact related to difficulties with interpersonal communication (Iverach et al., 2011). Research indicates that awareness of stuttering develops in children from two years of age (Ambrose and Yairi, 1994, Boey et al., 2009, Yairi, 1993). Further, typically fluent preschool-age children as young as 4 years of age tend to evaluate stuttered speech negatively (Ambrose & Yairi, 1994) and may react negatively towards preschool-age CWS in social interactions because of their stuttering (Langevin et al., 2009, Langevin et al., 2010). The early awareness of stuttering and other's negative reactions to stuttering likely explain the findings that CWS as young as the preschool-age tend to associate speaking with difficulty and exhibit more negative communication attitudes than CWNS (Clark et al., 2012; Guttormsen, Kefalianos, & Naess, 2015; Vanryckeghem, Brutten, & Hernandez, 2005). Perception of speaking as something that is difficult from such a young age may in turn adversely affect a child's ability to establish normally fluent speech-language planning and production. Furthermore, it may lay the foundation for the development of a negative cognitive bias, which is a significant risk factor for the development of anxiety later in life (Wong & Rapee, 2016). Negative communication attitudes in children who stutter may be an additional influential factor that could affect the level of autonomic arousal during speaking, as physiological responses (such as skin conductance and heart rate) were found to be strongly associated with cognitive bias in school-age children (e.g., Weems, Zakem, Costa, Cannon, & Watts, 2005). Accordingly, we hypothesize that preschool-age CWS who associate speaking with difficulty may display heightened levels of autonomic arousal during speaking.

Given the current multifactorial view of stuttering development, and the proposed roles of temperament and contextually-determined emotional arousal, it is important to determine the nature of speech-related autonomic arousal in preschool-age CWS and CWNS and its contributing factors. The nature of speech-related arousal in preschool-age children is also important to consider as autonomic arousal has been shown to affect speech motor control (Kleinow & Smith, 2006) and acoustic parameters of speech (Caruso et al., 1994; Arenas & Zebrowski, 2013). Heightened autonomic arousal may have a contributing role in the development of stuttering by affecting young children's emerging speech motor control skills and linguistic abilities (Smith & Weber, 2017; Arnold et al., 2014).

Thus, the purpose of the present study was to assess whether speech-related sympathetic nervous system arousal differed between preschool-age CWS and CWNS and whether it was associated with children's proclivity to experience negative emotions. We hypothesized that CWS will display heightened levels of autonomic arousal during speaking compared to CWNS. We further hypothesized that preschool-age children's proclivity for experiencing stronger and more frequent negative emotions will be associated with higher sympathetic nervous system activity during novel, potentially stressful speaking situations. Further, for CWS only, we examined whether their self-reported attitudes towards speaking had an effect on the level of speech-related sympathetic nervous system arousal. We hypothesized that preschool-age CWS who associate speaking with difficulty may display heightened levels of autonomic arousal during speaking. We employed a psychophysiological methodology to quantify speech-related arousal in preschool-age children who do and do not stutter.

The study addressed three specific questions:

(1) Do preschool-age CWS have a higher level of sympathetic nervous system arousal during speech production than CWNS, and does this depend on the speaking task?

(2) Do preschool-age CWS show greater negative affect than CWNS, and is negative affect associated with children's sympathetic nervous system arousal during novel speaking situations?

(3) Do preschool-age CWS show greater negative communication attitude than CWNS, and is communication attitude associated with CWS's sympathetic nervous system arousal during novel speaking situations?

Method

Thirty-two preschool-age children (age range: 36-67 months) and their caregivers participated in the study. Participants included 16 CWS (13 boys and 3 girls; mean age 3 years, 11 months; SD = 8.8 months) and 16 CWNS (12 boys and 4 girls; mean age 4 years, 1 month; SD = 9.9 months). All were paid volunteers recruited through an advertisement in a monthly parent magazine circulated throughout Syracuse 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 (Conture, 2001; Yaruss, 1998) (b) scored 11 or greater (i.e., severity of at least “mild”) on the SSI-4 (Riley, 2009), and (c) their caregivers expressed concern regarding stuttering. Stuttering severity of the CWS participants is presented in Table 1. No CWS had received treatment for stuttering prior to this study nor were they receiving any treatment at the time of the study. Children whose parents expressed no concern about their child’s fluency and who produced less than 3% stuttered disfluencies were assigned to CWNS group.

Procedures

All data collection procedures took place in the Syracuse University Stuttering Research Laboratory over two visits. During the first visit participants were administered standardized tests of speech and language and their caregivers responded to the study questionnaires. All psychophysiological data were collected during the second visit to the laboratory.

Speech, language and hearing abilities. All participants’ speech-language and hearing abilities were assessed using standardized measures. The “Sounds in Words” subtest of the Goldman-Fristoe Test of Articulation-2 (GFTA-2; Goldman & Fristoe, 2000) was used to assess children’s articulation skills. Receptive and expressive language abilities were evaluated using the Clinical Evaluation of Language Fundamentals – Preschool 2 (CELF-P2; Wiig, Secord, & Semel, 2005). Participants’ speech and language standard scores are presented in Table 2. In addition, 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).

Measure of temperament. Children's temperament was measured with the Children's Behavior Questionnaire Short Form (CBQ, Rothbart, Ahadi, Hershey, & Fisher, 2001; Putnam & Rothbart, 2006) which was administered to the caregiver (mothers in the majority of cases) who brought the child to the lab. The CBQ is a normed instrument with established validity and reliability that has been successfully used in other research on temperament and childhood stuttering (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Eggers, De Nil, & Van den Bergh, 2010). The CBQ short form 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 behavior 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).

Whereas the entire CBQ was administered to assess the participants' temperament, we were specifically interested in the CBQ factor of Negative Affectivity and the five behavior dimensions that contribute to this factor (anger/frustration, discomfort, fear, sadness, and soothability). This factor was chosen because it reflects a child's tendency to experience negative emotions, a temperamental quality found to be associated with development of chronic stuttering by Ambrose et al. (2015).

Measure of children's communication attitude. KiddyCAT (Vanryckeghem & Brutten, 2007) was administered to assess children's attitude towards own speech. The KiddyCAT is a twelve-item questionnaire, designed to obtain 3–6 year old children's self-reported attitude towards their speaking ability. The KiddyCAT has been extensively researched and shows good validity and reliability (Vanryckeghem and Brutten, 2007). The KiddyCAT requires children to agree/disagree with 12 statements describing their communication. The examiner reads aloud each of the 12 KiddyCAT statements to which children respond with ‘yes’ or ‘no’ indicating what they think about their speech. Scores for the 12 items are summed. A higher score on the KiddyCAT suggests greater negative attitudes towards one's speech. Additionally, Clark et al.’s (2012) factor analysis results suggested that a single dimension, namely speech difficulty, is reflected in the KiddyCAT questionnaire items.

Autonomic nervous system procedures and measures.

On the second visit to the laboratory, participants were seated at a table directly in front of a computer monitor. They first viewed an emotionally neutral animated screensaver of a three-dimensional fish tank for four minutes to establish a baseline level of electrodermal activity. After the baseline electrodermal activity was acquired, participants engaged in the two speaking tasks presented in the following order: (1) *Picture Description Task*; (2) *the Syllable Repetition Task* (SRT; Shriberg et al., 2009). These tasks were designed to elicit a range of speech-related autonomic reactivity as described below.

Speaking tasks. A *Picture Description Task* was chosen as a first speaking “stressor” as it resembles communicative speech production with an unfamiliar adult (the task often employed in psychophysiological and personality research with young children to evoke a social challenge). Importantly, it elicits narratives with a standardized context to allow for between-participant consistency. Participants were shown pictures from a wordless storybook about a boy, a dog, and a frog by the author Mercer Mayer, Frog Goes to Dinner (Mayer, 1974). To keep the narrative elicitation procedure consistent between the participants, the examiner was not allowed to ask specific questions about the picture but could only prompt the participant to tell them what was happening in the picture by saying "Let's look at this picture. Tell me what is happening here." The examiner was instructed to provide no more than three such elicitation prompts per picture. Narratives produced in response to the pictures were transcribed using the Systematic Analysis of Language Transcripts (SALT; Miller & Iglesias, 2008). Number of words and a mean length of utterances in morphemes for each participant's narrative were calculated using SALT. SALT-based written transcripts and acoustic analysis were also used to address separate research questions, not included in this report.

The Syllable Repetition Task (SRT; Shriberg, Lohmeier, Campbell, Dollaghan, Green, & Moore, 2009) was chosen as a second speaking "stressor" in the present study because non-word repetition tasks invoke a range of processes that underlie speech-language production such as auditory-perceptual, memory, and speech-language planning processes (Shriberg et al., 2009). Poor performance on these have been linked to the presence of developmental speech-language disorders (Bishop, 2002a, 2002b). Of particular relevance to the present study, is the fact that preschool-age CWS tend to have some difficulty with non-word repetition, resulting in overall lower non-word repetition accuracy than their normally fluent peers as reported in several research studies (Anderson, Wagovich, & Hall, 2006; Hakim & Bernstein-Ratner, 2004; Pelczarski & Yaruss, 2016) and summarized in a recent meta-analysis (Ofoe, Anderson, & Ntourou, 2018). Further, non-word repetition ability has been linked to stuttering persistence (Spencer & Weber-Fox, 2014).

The SRT was chosen among other non-word repetition tasks because it only includes voiced early-developing consonants (i.e., /b, d, m, n/) and one vowel (/ɑ/), sounds that will be in the phonemic inventories of young children, even those who have a speech sound disorder. Thus, this test minimizes confounds associated with misarticulations while still examining speech processing constraints. The administration and scoring procedures outlined in the SRT were followed (Shriberg et al., 2009). In brief, a standard digital version of the SRT was used to present non-words on a computer. The participants were told that they were going to hear a woman say some silly words on the computer and that they need to say each word exactly the way the woman says them. Following the scoring guidelines, deletions and substitutions of the target consonants were scored as incorrect. Sound distortions were scored as correct; it should be noted, however, that no participants in the present study produced any distortions of the four target consonants.

Sympathetic measures. Electrodermal activity and an acoustic signal were acquired simultaneously using the Biopac MP150 hardware system (Biopac Systems, Inc.) and recorded using Acknowledge software (ver. 4.3 for PC, Biopac). Electrodermal activity was recorded with electrodermal response transducers (model TSD 203) which included a set of two Ag-AgCl electrodes with incorporated molded housings designed for finger attachment. The response transducers were filled with an isotonic electrolyte gel and were placed on the volar surfaces of the middle phalanges of the index and middle fingers of the participants' right hand.

Standardized procedures for electrodermal activity recordings were implemented throughout all speaking tasks (Boucsein et al., 2012). The electrodes were connected to a Biopac GSR100C skin conductance amplifier. The electrodermal activity (expressed in microSiemens, μS) was sampled at 10 kHz with the gain set at 10 μS/V and a low-pass filter at 1 Hz and subsequently downsampled for the analysis.

The data were visually inspected during data collection to monitor for any instances of artifacts. In rare cases when participants pulled off the electrodes during the data collection resulting in intervals of missing data, the "Connect Endpoints" math function of the Biopac AcqKnowledge 4.3 software was then used to correct these artifacts. No more than five percent of the total data for any one condition (baseline or speaking) were corrected using this procedure. To measure tonic arousal, mean SCL and number of non-specific SCRs (i.e., spontaneous fluctuations in electrodermal activity) were calculated for the baseline and the Picture Description task using AcqKnowledge 4.3 software from a continuous electrodermal activity signal. Following common procedures (e.g., Boucsein et al., 2012) SCL was calculated after phasic responses were removed from the signal. To be able to compare non-specific SCRs across narratives of different lengths, frequency rather than the number of non-specific SCRs was chosen for the analysis. The frequency of non-specific SCRs was calculated as the number of responses per minute.

For the *Syllable Repetition Task* in addition to the mean SCL, we calculated the number of specific SCRs elicited by the non-words. These specific SCRs were time-locked to the presentation (i.e., onset of audio recording) of each of the non-words during the *Syllable Repetition Task*. The time window for specific SCR latency was set from 1 to 4 seconds following recommendations of Boucsein et al. (2012).

Description of dependent variables. For the *Picture Description Task*, a SCL residualized change score (see explanation below) and frequency of non-specific SCRs served as the dependent variables. For the *Syllable Repetition Task*, a SCL residualized change score and a number of specific SCRs elicited by the *Syllable Repetition Task* non-words were the dependent variables.

The law of initial values (Wilder, 1958) suggests that baseline SCL values could influence SCL in other experimental conditions. For the first research question, we entered baseline SCL as a covariate in the model to control for its effect on the SCL in the speaking tasks, and calculated SCL residualized change scores (Llabre, Spitzer, Saab, Ironson, & Schneiderman, 1991; Jones et al. 2014; Zengin-Bolatkale, Conture & Walden, 2015) for both speaking tasks. These residualized SCL change scores served as the dependent variables in the subsequent analyses.

Statistical Analyses

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 measures (mean, standard deviation, variance, skewness and kurtosis).

Univariate general linear models (with repeated measures on the speaking tasks) were performed for the analyses with SCL or frequency of non-specific SCRs as the dependent variables. Due to the non-normal distribution of SCRs, univariate generalized linear models that allowed for skewed distributions (Nelder & Wedderburn, 1972) were performed when the number of specific SCRs was the dependent variable. An alpha level was set at $p \leq 0.05$ 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.

Group differences on possible confounding variables

Due to the potential influence of speech-language skills and age on sympathetic arousal during speech, we examined whether CWS and CWNS groups had significant differences in those variables.

A multivariate analysis of variance (ANOVA) revealed no significant between-group differences in chronological age, CELF-P2 Core Language and mean length of utterances produced during the Picture Description Task (measured in morphemes; see Table 2). However, CWNS had a higher standard score on the GFTA-2 ($F_{1,30}$ = 6.448, p = .017) and CWNS also produced significantly more words during the Picture Description Task than CWS ($F_{1,30}$ = 5.912, p = .021).

A linear mixed effects model (with repeated measures on the *Syllable Repetition Task* non-word length) indicated that CWS had a significantly lower repetition accuracy on *Syllable Repetition Task* than CWNS at all syllable lengths ($F_{1,30}$ = 5.468, p = .026): two-syllable non-words (t = 4.55, p = 0.032, β = 9.44) three-syllable non-word length (t = 2.12, p = .043, β = 14.38) and four-syllable non-words (t = 2.11, p = .043, β = 14.88). For that reason, we included repetition accuracy as an independent variable in the model addressing Research Question 1. It should be noted that accuracy errors did not include any instances of stuttering. All children in the CWS group were able to repeat the non-words fluently. Means and SDs for non-word repetition accuracy are reported in Table 3.

Research Question 1: Do preschool-age CWS have a higher level of sympathetic nervous system arousal during speech production than CWNS, and does this depend on the speaking task?

Univariate general linear model (with repeated measures on the speaking tasks) revealed no significant group differences in SCL during the two speaking tasks ($F_{1,29}$ = 1.439, p = .240, η^2_p = .047). As expected, there was a significant main effect of baseline SCL on the SCL during the speaking tasks ($F_{1,29}$ = 26.866, p < .0001, η^2_p = .481). Both groups showed an increase in SCL from baseline to the speaking tasks (see Figure 1). The model also indicated a marginally significant effect of the speaking task ($F_{1,29}$ = 3.891, p = .058, η^2_p = .118), with both groups showing a greater increase in SCL during the *Syllable Repetition Task* (*β = 1.121*) than during the *Picture Description Task* (*β = .998)* compared to the baseline SCL. Although the group effect was not significant, a standardized regression coefficient beta (i.e., *β)* indicated a trend of larger increases in SCL for CWNS than CWS during the speaking tasks (for CWNS, *β = .952* in the *Syllable Repetition Task* and *β = .846* in the *Picture Description Task*). Univariate general linear model revealed no group difference in the frequency of non-specific SCRs during the picture description task ($F_{1,30}$ = .167, p = .686, η^2_p = .006; see Figure 2). Additionally, there was no significant difference between CWS and CWNS in the baseline SCL ($F_{1,30}$ = .152, p = .699, η^2_p = .005; *β* = -*.262* for CWNS group).

Univariate generalized linear model revealed a significant effect of Group (Wald χ^2 = 3.836, df = 1, p = .050, *β* = 4.8), and Group x Repetition Accuracy interaction (Wald χ^2 = 4.343, df = 1, p = .037, *β* = -.062) for the number of specific SCRs elicited during the *Syllable Repetition Task*. CWS produced more specific SCRs in response to non-words on the *Syllable Repetition Task* than CWNS (see Figure 3). To follow up on the interaction effect, two univariate generalized linear models were fit to each group's data. These analyses indicated no significant effect of Repetition Accuracy on the number of specific SCRs produced by either CWS (Wald χ^2 = 1.784, df = 1, p = .182, *β* = .028) or CWNS (Wald χ^2 = 2.582, df = 1, p = .108, *β* = -.034). However, a standardized regression coefficient beta (i.e., *β)* indicated that the association between Repetition Accuracy and the number of specific SCRs was in the opposite direction within the groups. For CWS, a higher Repetition Accuracy was associated with a greater number of specific SCRs. Conversely, for CWNS, a higher Repetition Accuracy was associated with fewer specific SCRs (see Figure 4).

Research Question 2: Do preschool-age CWS show greater negative affect than CWNS and is negative affect associated with children's sympathetic nervous system arousal during novel speaking situations?

A univariate ANOVA revealed no significant between-group difference on the CBQ Negative Affectivity factor score. However, a multivariate ANOVA for the 5 individual scale scores that yield the Negative Affectivity composite score revealed significant between-group differences in the Fear scale ($F_{1,30}$ = 4.42, p = .044) and the Sadness scale ($F_{1,30}$ = 5.69, p = .024). Caregivers rated CWS higher on the Fear and Sadness scales. Means and SDs for CBQ scores are reported in Table 4.

Univariate general linear model (with repeated measures on the speaking tasks) revealed neither a significant main effect of CBQ Negative Affectivity score on SCL during the speaking tasks ($F_{1,29}$ = 2.84, p = .102, η^2_p = .089), nor a significant effect of Group x CBQ Negative Affectivity interaction ($F_{1,29}$ = .749, p = .394, η^2_p = .025).

There was no significant effect of either CBQ Negative Affectivity score ($F_{1,29}$ = 2.672, p = .113, η^2_p = .084) or Group x CBQ Negative Affectivity interaction ($F_{1,29}$ = .001, p = .973, η^2_p < .001) on the frequency of non-specific SCRs during the Picture Description Task. Further, there was no significant effect of either CBQ Negative Affectivity score (Wald χ^2 = .447, df = 1, p = .504, β = -.200) or Group x CBQ Negative Affectivity interaction (Wald χ^2 = .344, df = 1, p = .557, β = -.066) on the number of specific SCRs elicited by the *Syllable Repetition Task* non-words.

Research Question 3: Do preschool-age CWS show greater negative communication attitude than CWNS, and is it associated with their sympathetic nervous system arousal during novel speaking situations?

A univariate ANOVA revealed a significant group difference in KiddyCAT questionnaire scores ($F_{1,27}$ = 7.507, p = .011, β = -1.75), with CWS scoring higher than CWNS (means and SDs are reported in Table 1). Three children (all three were in the CWNS group) were excluded from this analysis because they were not able to reliably respond to KiddyCAT questions.

Univariate general linear model (with repeated measures on the speaking tasks) revealed no significant effect of the KiddyCAT score on CWS's SCL during the speaking tasks ($F_{1,14}$ = 1.080, p = .316, η^2_p = .072). Although this was a non-significant trend, a standardized regression coefficient beta indicated that a higher KiddyCAT score was associated with a larger increase in SCL during speaking (β = .152 for the Picture Description Task; β = .119 for the *Syllable Repetition* Task). There was no effect of the KiddyCAT score on the frequency of non-specific SCRs during the *Picture Description Task* ($F_{1,14}$ = .032, p = .862, η^2_p = .002, β = -.048) or on the number of specific SCRs elicited by the non-words during the *Syllable Repetition Task* (Wald χ^2 = 0.434, df = 1, p = 0.510, β = -0.105).

Discussion

The present study resulted in three main findings. First, preschool-age CWS did not differ from CWNS peers in their level of sympathetic arousal during the Picture Description Task. However, during the *Syllable Repetition Task*, preschool-age CWS had a higher sympathetic arousal level than their CWNS peers. Second, preschool-age CWS were rated by their caregivers as more fearful and prone to sadness. However, preschool-age CWS and CWNS's tendency to experience stronger and more frequent negative emotions was not associated with their sympathetic arousal during the speaking tasks. Third, for preschool-age CWS, negative communication attitude was not associated with the level of sympathetic arousal during speaking. The implications of these findings are discussed below.

Differences in Speech-related Sympathetic Arousal of preschool-age CWS and CWNS

Previous research indicates that preschool-age CWS do not demonstrate greater sympathetic arousal during such speech tasks such as picture descriptions (Choi et al., 2016) or picture naming (Zengin-Bolatkale et al., 2015) compared to CWNS peers. Additionally, voice measures of reactivity to speech tasks (fundamental frequency of voice) assessed in preschool-age CWS and CWNS also indicate no difference in speech-related arousal (Kazenski et al., 2014). Consistent with these findings, the present study indicated that preschool-age CWS did not differ from CWNS peers in their level of sympathetic arousal during the Picture Description Task. Based on the previous work, we hypothesized that, for CWS, a narrative task, such as a picture description, may be more challenging than for CWNS and might elicit a higher sympathetic arousal level. The Picture Description Task in our study may have been relatively easy and not as taxing for preschool-age children (and CWS specifically) as we hypothesized. Specifically, the hypothesized stress from speaking with an unfamiliar adult may not have occurred for our participants as expected. Our finding that the Picture Description Task elicited a lower level of sympathetic arousal compared to the *Syllable Repetition Task* supports this interpretation. Of note, consistent with previous research in adults (Peters & Hulstijn, 1984; Weber & Smith, 1990) and children who stutter (Arnold, MacPherson, & Smith, 2014; Choi et al., 2016) we found that both speaking tasks were associated with an increased sympathetic activation compared to the baseline SCL.

Consistent with our hypothesis, preschool-age CWS demonstrated greater sympathetic arousal during the *Syllable Repetition Task* than their CWNS peers. This finding suggests that the non-word repetition task taxed the cognitive-linguistic system of both groups of children, particularly that of preschool-age CWS, thus resulting in greater autonomic arousal. Two other findings from this study lend support to this interpretation. First, the *Syllable Repetition Task* elicited a trend toward greater sympathetic arousal from both groups of participants compared to the *Picture Description Task*. Second, CWS had lower accuracy on the *Syllable Repetition Task* compared to CWNS. Moreover, for CWS, a higher repetition accuracy was associated with a greater number of specific SCRs. Conversely, for CWNS, a higher repetition accuracy was associated with fewer specific SCRs. As reviewed in a recent meta-analysis study (Ofoe, Anderson, & Ntourou, 2018), published research suggests that non-word repetition represents an area of weakness for preschool-age CWS. Accordingly, higher level of sympathetic arousal can be indicative of a greater effort that CWS exerted during the task, as cognitive effort has been robustly associated with increased sympathetic arousal (for review see Boucsein, 2012). It should be noted that disfluency did not contribute to CWS's lower performance on the SRT as the participants repeated all non-words fluently.

Non-word repetition invokes a range of processes that underlie speech-language production. Presently, it is not clear what specific process involved in non-word repetition invokes the difficulty for preschool-age CWS. As reviewed in a recent study, CWS's difficulties could be a result of lower auditory-perceptual skills, phonetic encoding, reduced verbal short-term memory, and/or speech planning and execution processes (Anderson, Wagovich, Brown, 2019). This study's design does not allow us to differentiate which process contributed to the CWS's difficulty with SRT. Others, however, have suggested that phonological working memory skills may be implicated in CWS's performance (Hakim &Ratner, 2004; Anderson, Wagovich, Hall, 2006). This study adds to the evidence that non-word repetition may be more difficult for CWS.

Impact of Negative emotional reactivity on Speech-related Sympathetic Arousal

Previous research on temperamental qualities of CWS using parent-report questionnaires (e.g., CBQ) indicates that preschool-age CWS tend to experience negative emotions with higher frequency and intensity than CWNS peers. Among these negative emotions, higher scores on the Fear index differentiated preschool-age CWS from CWNS in two large studies (Ambrose et al., 2015; Eggers, De Nil, & Van den Bergh, 2010). Consistent with previous literature, we found that caregivers of CWS in our study rated their children as more fearful compared to CWNS. We also found that caregivers of CWS rated their children higher on CBQ Sadness scale than caregivers of CWNS. Given that the children's tendency to experience stronger negative emotion is a risk factor for the development of anxiety disorder later in life (Cote et al., 2009), children who stutter and have a proclivity to experience stronger negative emotions (specifically fear) may be at an increased risk for developing an anxiety disorder later in life. Further study of this hypothesized association is warranted, especially given the evidence that the proclivity to experience stronger and more frequent negative emotions was associated with the development of chronic stuttering in children (Ambrose et al., 2015).

Based on the published data indicating an association between sympathetic nervous system activity during various challenging tasks and personality in preschool-age children (e.g., Fowles, Kochanska, and Murray, 2000; Nikolić, Aktar, Bögels, Colonnesi, & de Vente, 2018, cf. Alkozei, Creswell, Cooper, & Allen, 2015) we hypothesized that there would be an association between preschool-age children's tendency to experience negative emotions and their level of sympathetic arousal during speaking. Our data did not support this hypothesis. Our finding, however, corroborates the finding of Choi et al. (2016) who also did not find a significant association between preschool-age CWS's negative emotional reactivity and their SCL during a similar picture description task. Research suggests that sympathetic arousal is highly dependent on the stimulus characteristics (e.g., Mardaga & Hansenne, 2010), thus two factors should be considered for our results interpretation. Our study was not designed to elicit any emotional response from the participants either before or during the speaking tasks. The social stress of the speaking tasks may also have been lowered as the participants engaged in the speaking tasks on their second visit to the laboratory and were somewhat familiar with the environment and research staff. Thus, future studies should attempt to elicit an emotional response and/or increase the social stress of the speaking tasks to test whether this might reveal the hypothesized association between personality and sympathetic nervous system activity in preschool-age children.

Communication Attitude and its Impact on Speech-related Sympathetic Arousal for CWS

Prior research indicates that CWS as young as three years of age experience more negative attitudes towards speech than CWNS, which is consistent with our finding that preschool-age CWS gave higher scores on the KiddyCAT questionnaire indicating a more negative communication attitude compared to their CWNS peers. Contrary to our hypothesis, however, there was no significant effect of the KiddyCAT score on CWS's level of sympathetic arousal during the speaking tasks. Although our data suggest that preschool-age CWS have experienced some difficulties with communication (as reflected in their higher KiddyCAT scores) it is possible that preschool-age CWS, who are close to the onset of stuttering, have not had sufficient negative or frustrating speech-related experiences in social situations that may lead to an increased emotional arousal and sympathetic activation during a conversation. Research findings that communication attitudes of CWS tend to worsen with age (De Nil & Brutten, 1990; Vanryckeghem et al., 2005) support this interpretation. Our findings are also similar to those of van der Merwe et al. (2011), who assessed speaking-related state and trait anxiety in preschool-age CWS and CWNS and found no difference between the groups on parent-report measures of anxiety and cortisol levels prior to and after engaging in a conversation with an examiner. Together these findings suggest that although preschool-age CWS experience some communication difficulties or apprehension, these experiences may be context-specific and not pervasive enough to result in increased sympathetic arousal in all communication interactions.

Caveats

We acknowledge several limitations of this study. First, as described in the Method section the groups were not matched on gender: CWS group included 13 boys and 3 girls, whereas CWNS group included 12 boys and 4 girls. Although the gender imbalance between the groups is minimal, it could have affected the results.

Second, our protocol did not allow us to differentiate specific SCRs elicited by 2-syllable SRT non-words compared to those elicited by 3- or 4-syllable SRT non-words. It is possible that longer non-words elicit more SCRs than shorter non-words. As higher linguistic complexity may be associated with higher sympathetic arousal, the present study might serve to motivate future study of the association between sympathetic arousal and non-word length.

Third, the two speaking tasks employed in the study were presented in the same order for all participants (the *Picture Description Task* first, the *Syllable Repetition Task* second). The order of task presentation was decided based on the anticipated difficulty of the tasks for our preschool-age participants (similar to other studies e.g., Arnold et al., 2014). The fact that we did not counterbalance the order of the two experimental tasks could have contributed to our finding that the Picture Description Task elicited a lower level of sympathetic arousal compared to the *Syllable Repetition Task.*

Conclusions

Our findings suggest that age-appropriate social communication tasks are not inherently more stressful for preschool-age children who stutter and not associated with state-related stress or anxiety that is often reported for adults who stutter. However, speaking tasks that place a higher demand on children's cognitive-linguistic system may be more taxing and challenging to preschool CWS, leading to a higher level of arousal.

More negative communication attitude was evidenced in preschool-age CWS compared to CWNS peers. Thus, taking prior research into perspective, it appears that at the onset of stuttering, preschool-age CWS may already have experienced some difficulties with communication. Existing research with older children and adults who stutter suggests that these early difficulties are likely to worsen as the negative social-emotional impact of stuttering becomes greater over time, with the increased age and longer history of stuttering.

Lastly, consistent with previous literature, we found that caregivers of CWS in our study rated their children as more fearful compared to CWNS. Given the reported associations between a proclivity to experience negative emotions and development of chronic stuttering on one hand and development of anxiety on the other hand, further study of the role of temperament in stuttering development is warranted.

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Table 1: Stuttering severity, assessed by the Stuttering Severity Instrument - 4 (SSI-4; Riley, 2009) for children who stutter (CWS).

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

Table 2: Speech, language and communication attitude scores for children with do (CWS, n = 16) and do not stutter (CWNS, n = 16).

Independent variable	Group	Mean	Std. Deviation	Difference Significant
Age (months)	CWNS	49.63	9.90	*n.s.*
	CWS	46.56	8.82	
GFTA Standard Score	CWNS	104.63	12.10	$p = 0.017$
	CWS	92.75	14.30	
CELF-P2 Core Language Standard Score	CWNS	112.63	12.34	*n.s.*
	CWS	105.38	11.11	
Mean Length of Utterances during picture description	CWNS	5.59	1.79	*n.s*
	CWS	4.92	1.75	
Number of Words Spoken during picture description	CWNS	409.69	130.89	$p = 0.021$
	CWS	288.31	150.79	
KiddyCAT score	CWNS	2	1.528	$p = 0.011$
	CWS	3.75	1.844	

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

Table 3: The Syllable Repetition Task Accuracy scores* for children with do (CWS, n = 16) and do not stutter (CWNS, n = 16).

Independent variable	Group	Mean	Std. Deviation	Difference Significant
Percent consonants correct at 2-syllable level	CWNS	95.81	8.328	*p* = 0.032
	CWS	86.38	14.573	
Percent consonants correct at 3-syllable level	CWNS	86.69	15.928	*p* = 0.043
	CWS	72.31	21.981	
Percent consonants correct at 4-syllable level	CWNS	78.44	19.517	*p* = 0.043
	CWS	63.56	20.298	

Note: Accuracy was measured in percent of consonants correctly produced.

Table 4: CBQ scores for the composite factor of Negative Affectivity and the associated subfactor (individual scale) scores for children with do (CWS, n = 16) and do not stutter (CWNS, n = 16).

Temperamental quality	Group	Mean	Std. Deviation	Difference Significant
Negative Affectivity Factor	CWNS	3.683	.667	*n.s*
	CWS	4.050	.663	
Anger/frustration scale	CWNS	4.223	1.109	*n.s*
	CWS	4.238	1.079	
Discomfort scale	CWNS	3.958	1.065	*n.s*
	CWS	4.104	.925	
Fear scale	CWNS	3.365	.908	$p = 0.044$
	CWS	4.200	1.306	
Sadness scale	CWNS	3.880	1.026	$p = 0.024$
	CWS	4.618	.693	
Soothability scale	CWNS	5.010	.754	*n.s*
	CWS	4.901	.964	

Note: *n.s.* = not significant

Figure 1: Mean skin conductance levels (SCL) in baseline and speaking tasks for children who do (CWS, n = 16) and do not stutter (CWNS, n = 16).

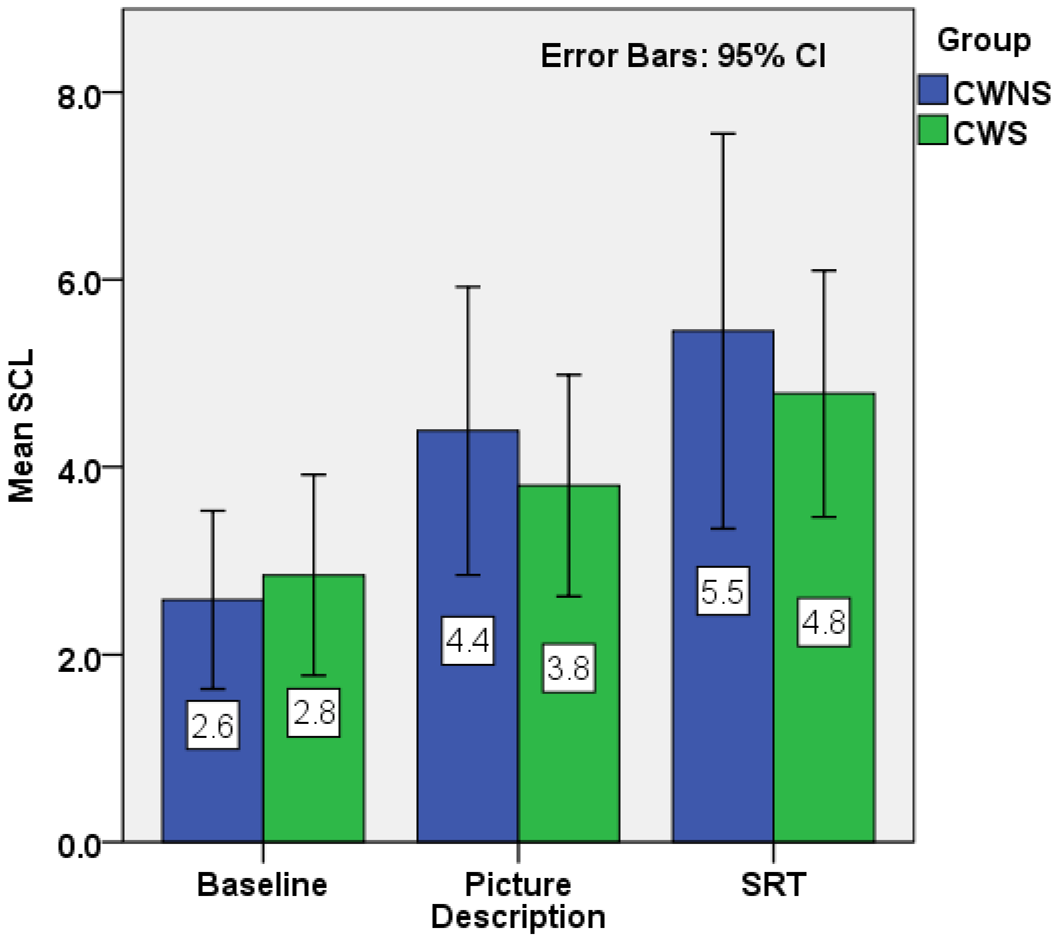

Figure 2: Mean frequency of non-specific skin conductance responses during the Picture Description task for children who do (CWS, n = 16) and do not stutter (CWNS, n = 16).

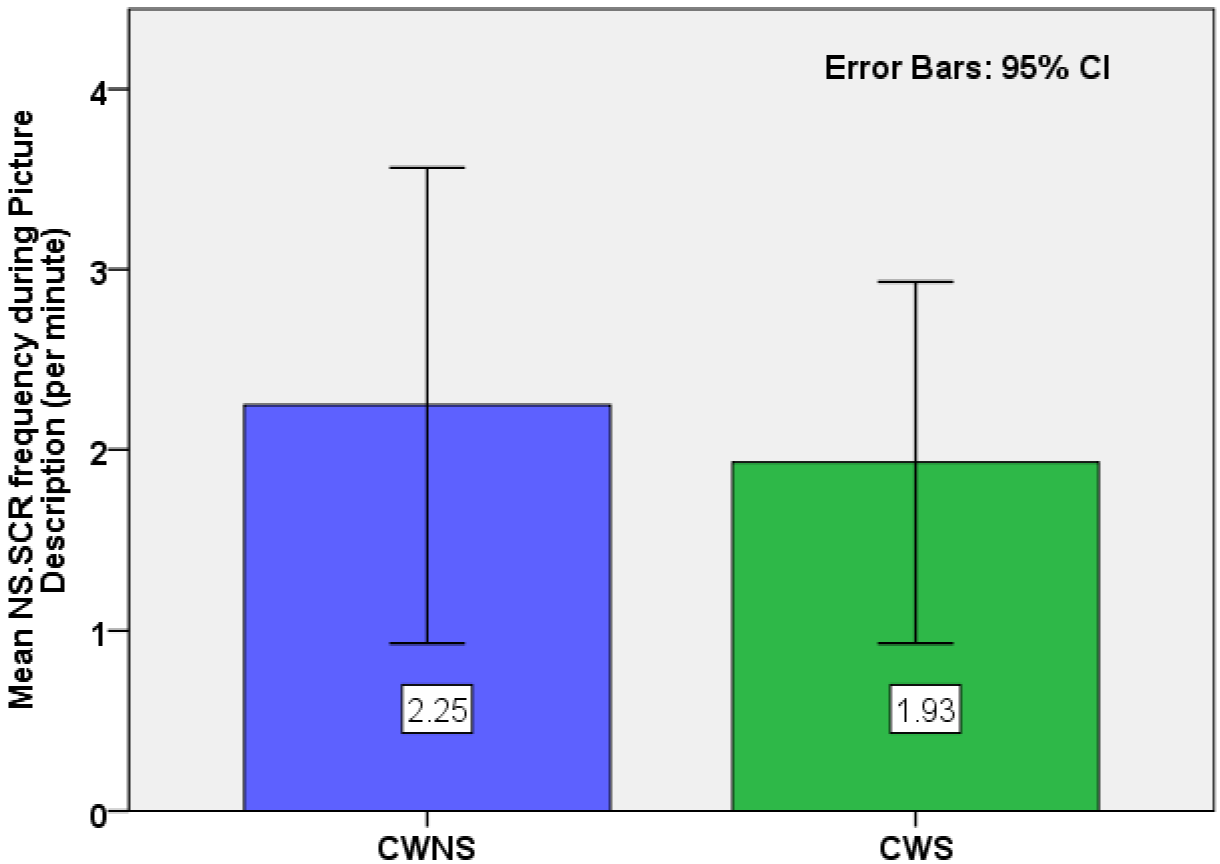

Figure 3: Histogram for the number of specific SCRs elicited by the SRT non-words in children who do (CWS, n = 16) and do not stutter (CWNS, n = 16).

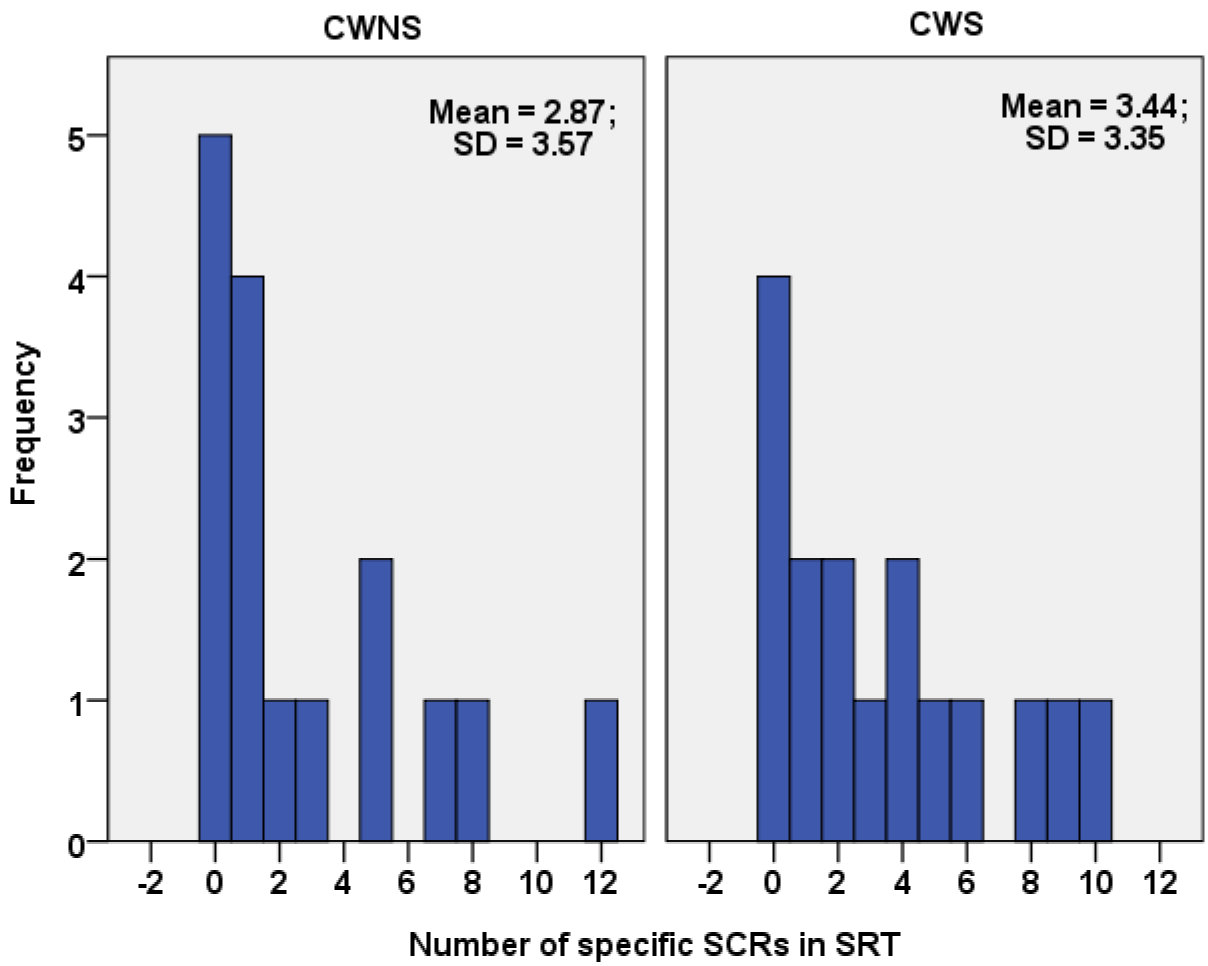

Figure 4: Association between repetition accuracy (percent of consonants correctly produced) and a number of specific SCRs elicited by the SRT non-words for children who do (CWS, n = 16) and do not stutter (CWNS, n = 16).