Research Article

Physiological Correlates of Fluent and Stuttered Speech Production in Preschool Children Who Stutter

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Purpose: The purpose of this study was to compare physiological indices of sympathetic nervous system arousal recorded during fluent and stuttered utterances in a preschool children who stutter (CWS).

Method: Twenty-two 4- to 5-year-old CWS participated in the experiment. We recorded children's skin conductance response amplitude and frequency, blood pulse volume amplitude, and pulse rate as they completed a picture description task. We then compared indices of phasic sympathetic arousal recorded during stuttered versus fluent utterances. In addition, children's communication attitudes were evaluated with a self-report measure.

Results: We detected significantly higher sympathetic arousal during stuttered utterances compared to fluent utterances. Specifically, we found larger skin conductance responses occurring at an increased frequency and decreased blood pulse volume amplitudes during stuttered speech. The behavioral measure indicated a negative communication attitude in only one-third of the participants.

Conclusion: Our findings suggest that preschool CWS may exhibit higher levels of sympathetic arousal during stuttered speech compared to when they are speaking fluently. We discuss the potential impact of increased sympathetic arousal on speech regulatory mechanisms in early childhood stuttering and present questions to guide future research.

Theories and anecdotal accounts implicate emotional factors in stuttering, although the precise role that these factors play in the onset and progression of the disorder is unclear (Sheehan, 1953; A. Smith & Weber, 2017; Van Riper, 1972; Walden et al., 2012; Zimmermann, 1980). Walden et al.'s dual diathesis–stressor account of stuttering posits that high emotional reactivity and reduced self-regulatory mechanisms contribute to the frequency of stuttering-like disfluencies (SLDs; Walden et al., 2012). In the demands and capacities model of stuttering, the demands of challenging speaking circumstances or environments may breach an intrinsic threshold or capacity for fluent speech production (Adams, 1990; Starkweather & Gottwald, 1990). Clearly, the multifactorial nature of childhood stuttering suggests that a propensity toward heightened anxiety or a decreased ability to self-regulate alone is not a univariate

cause of the disorder. In their multifactorial dynamic pathways theory, A. Smith and Weber (2017) propose that childhood stuttering results from an impairment in speech sensorimotor control conditioned by the interaction of emotional, motoric, linguistic, and cognitive factors.

Previous investigations into the relationship between emotional factors and stuttering in adults have focused on “trait” anxiety, or the inherent propensity to be anxious, as well as “state” anxiety arising during particular circumstances, namely, social interactions (Connally et al., 2018; Craig, Hancock, Tran, & Craig, 2003; Mulcahy, Hennessey, Beilby, & Byrnes, 2008). A meta-analysis of anxiety research in stuttering over the last four decades confirmed that, for many adults who stutter (AWS), social/communicative anxiety is a significant problem (Craig & Tran, 2014).

Research examining the role of emotional processes in early childhood stuttering has explored differences in relatively stable trait constructs such as temperament, including emotional reactivity and emotional regulation between children who stutter (CWS) and children who do not stutter (CWNS; see the review in Jones, Choi, Conture, & Walden, 2014). Evidence from behavioral observation, caretaker reporting, and psychophysiological measures suggest that differences in emotional reactivity and emotion

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regulation are associated with childhood stuttering. Several studies found group differences between preschool CWS and CWNS in their state of emotional reactivity and regulation during experiments designed to induce arousal using observational measures (K. N. Johnson, Walden, Conture, & Karrass, 2010; Jones, Conture, & Walden, 2014; Ntourou, Conture, & Walden, 2013; Walden et al., 2012). For example, preschool-age CWS displayed significantly more negative emotional expressions than CWNS after receiving a disappointing gift (K. N. Johnson et al., 2010). Temperamental traits of preschool CWS are typically assessed through parent report measures such as the Behavioral Style Questionnaire (McDevitt & Carey, 1978) or the Children's Behavior Questionnaire (Rothbart, Ahadi, Hershey, & Fisher, 2001). Compared to CWNS, several studies have found temperamental differences in some CWS, such as increased anger or frustration, and greater difficulty regulating emotions or adapting to change (Anderson, Pellowski, Conture, & Kelly, 2003; Eggers, De Nil, & Van den Bergh, 2010; Karrass et al., 2006). However, others' and our research have not revealed temperamental differences between young CWS and CWNS using parent report measures (Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014, 2017; Walsh, Smith, Christ & Weber, 2019).

Psychophysiological Investigations in AWS and CWS

A growing number of studies have used indices of autonomic nervous system (ANS) activation to provide a physiological correlate of emotion in preschool CWS and CWNS (Choi, Conture, Walden, Jones, & Kim, 2016; Jones, Buhr, et al., 2014; Jones et al., 2017; Zengin-Bolat kale, Conture, & Walden, 2015; Zengin-Bolat kale, Conture, Walden, & Jones, 2018). ANS measures allow for the examination of covert and often nonconscious physiological arousal associated with emotional, cognitive, and motoric processes in CWS. The sympathetic and parasympathetic branches of the ANS dynamically modulate internal physiological functions to maintain homeostasis, helping us adapt to internal and environmental demands. The ANS is a mediator between brain and body; bidirectional pathways transmit efferent signals to the periphery while information regarding visceral states is relayed back to the central nervous system influencing our thoughts, emotions, and actions (Cardinali, 2018; Jänig, 2006). Efferent signals from the sympathetic nervous system (SNS) can trigger multiple simultaneous systemic responses—accelerated heart rate, blood vessel constriction, and increased perspiration, accounting for the all-encompassing physiological response to stressors (Andreassi, 2000). Electrodermal measures, such as skin conductance level (SCL) and skin conductance response (SCR), and blood pulse measures are used to quantify activation of the SNS (Andreassi, 2000). SCL is a tonic measure of electrodermal activity recorded over longer periods and is not specifically associated with the onset of a stimulus. Alternatively, SCRs are phasic changes in sympathetic arousal that occur within a second

or two of a specific stimulus (Mendes, 2009). The amplitude and frequency of SCRs are associated with increased sympathetic arousal elicited by diverse psychological states (Dawson, Schell, & Filion, 2007). A decrease in blood pulse volume (BPV) amplitude is accomplished through vasoconstriction and indicates SNS arousal, while increases in pulse rate, a correlate of heart rate, are also associated with increased SNS arousal (Andreassi, 2000). A range of emotional states such as fear, joy, anger, and stress reliably engender increases in SNS arousal (Kreibig, 2010). Increased SNS activation is also associated with different cognitive states, for example, in response to novel stimuli (Bradley, 2009), under greater task demands (Mackersie & Calderon-Moultrie, 2016; Tulen, Moleman, van Steenis, & Boomsma, 1989), or with increased attention (Iani, Gopher, & Lavie, 2004).

Zimmermann (1980) initially proposed a link between ANS arousal and stuttering through the direct effects of the SNS on speech sensorimotor control. Subsequent investigations examining autonomic arousal in adult speakers did not, however, find appreciable differences in SNS arousal between AWS and typically fluent adults during speaking and other tasks (Peters & Hulstijn, 1984; Weber & Smith, 1990; Zhang, Kalinowski, Saltuklaroglu, & Hudock, 2010). More recently, Walden et al. conducted studies using ANS measures to assess emotional reactivity and regulation between groups of preschool CWS and CWNS during emotionally salient experimental tasks. One study found significantly higher SCLs in 3-year-old CWS compared to age-matched CWNS during a stress-inducing rapid naming task (Zengin-Bolat kale et al., 2015); however, group differences were not seen once children reached 4–5 years old. Jones, Buhr, et al. (2014) reported a group-by-condition effect in which CWS had higher SCLs while viewing a positively valenced video clip, while CWNS had higher SCLs while viewing a negative video clip. They also found that preschool CWS had higher SCLs compared to CWNS while producing a narrative after viewing a positive video clip. These studies suggest possible differences between CWS and CWNS in sympathetic activity during emotionally arousing tasks.

Awareness of Stuttering in Preschool Children

Yairi and Ambrose (2005) note that awareness of stuttering should logically precede negative emotions or anxiety toward stuttering. Awareness of stuttering begins to emerge in early childhood; however, documenting precisely a child's level of awareness is challenging. In their study of stuttering self-awareness, Ambrose and Yairi (1994) had groups of CWS and CWNS discriminate between two puppets, one that spoke fluently and the other who stuttered, by pointing to the puppet that “talks the way you do.” They found that only 15% of CWS consistently selected the disfluent puppet. Given the difficulty of the task, however, the authors acknowledged that their results may not capture self-awareness, especially in younger CWS. When more nuanced nonverbal (e.g., sighing, displays

of frustration) and verbal (e.g., remarks about stuttering, asking for help) indicators were considered, a parent report study of over 800 preschool CWS noted higher percentages of stuttering awareness in preschool CWS that increased over time from 65% of 3-year-old CWS to 80% of 5-year-old CWS (Boey et al., 2009).

Growing awareness of speech struggles is accompanied by negative reactions and self-perception of communication ability in some preschool CWS (e.g., Clark, Conture, Frankel, & Walden, 2012; Jones, Conture, & Walden, 2014; Vanryckeghem & Brutten, 1997). Guttormsen, Kefalianos, and Næss (2015) conducted a meta-analysis of 18 studies of communication attitudes in CWS and CWNS aged 3–18 years. They found that preschool CWS, on average, exhibited a more negative communication attitude compared to preschool CWNS, although this trend became more apparent as children grew older. Using observational measures, Jones, Conture, and Walden (2014) noted that preschool CWS demonstrated more negative displays of emotion prior to and during stuttered utterances compared to fluent utterances after they were exposed to a conversation with positive undertones.

Unlike the use of behavioral questionnaires and observation, SNS measures allow us to examine covert physiological responses during stuttering behaviors, offering an alternative perspective. Choi et al. (2016) had 47 CWS produce narratives to wordless picture books while they recorded SCL and computed percentage of SLDs. They did not find a correlation between SCL and stuttering severity, a finding they interpreted as not supporting the hypothesis that tonic skin conductance is a mediating variable in stuttering frequency. On the other hand, Weber and Smith (1990) reported that AWS had higher phasic SNS activity associated with stuttered speech compared to fluent speech produced during reading and spontaneous speaking tasks. As these adults had been stuttering since childhood, higher levels of arousal detected during stuttering may be a conditioned response to their disfluencies. To our knowledge, no study has explored levels of physiological arousal during fluent and stuttered speech in preschool CWS. This research is relevant to the role of SNS arousal in speech execution in early childhood stuttering.

Purpose of the Current Study

The aim of this study is to examine the relationship between SNS activity and speech production in preschool CWS. We compared indices of phasic SNS arousal during stuttered versus fluent utterances produced during a picture description task. We hypothesized that higher levels of SNS arousal would be associated with stuttered speech compared to fluent speech, given that awareness of stuttering is emerging in preschool CWS (Boey et al., 2009) and some CWS at this age may already display negative feelings toward communication (Guttormsen et al., 2015). As such, we also explored the potential relationships among stuttering severity, communication attitude, and SNS

indices to examine the premise that children who exhibit a more negative communication attitude or more severe stuttering may show heightened SNS arousal during stuttered utterances.

Method

Participants

The experimental protocol was approved by the Institutional Review Board at Purdue University and adhered to the Human Research Protection Program regulations and guidelines. We obtained written informed consent from parents or legal guardians at the beginning of the experiment. Data from 22 preschool CWS (19 boys and three girls; $M = 53.3$ months, $SD = 6.15$, range: 46–71) were included in this study. These children participated in a larger investigation conducted by the Purdue Stuttering Project comparing behavioral indices of temperament and SNS responses during speech and oral motor tasks in preschool CWS and their fluent peers (Walsh et al., 2019). We included children from the larger study in the current study if they produced at least six stuttered and six fluent utterances during a picture description task, described below.

Per parent report, all children were native speakers of North American English with no history of developmental, cardiovascular, or neurological disorders and normal or corrected-to-normal vision. Participants were screened for medications affecting the central nervous and cardiovascular systems (depressants, stimulants, analgesics, anti-coagulants, etc.) and had not consumed caffeine prior to the experiment per parent report. The children scored within normal limits on assessments of nonverbal intelligence (Primary Test of Nonverbal Intelligence; Ehrler & McGhee, 2008) and social development (Childhood Autism Rating Scale–Second Edition; Schopler, Van Bourgondien, Wellman, & Love, 2010) and passed a bilateral pure-tone hearing screening at 500, 1000, 2000, 4000, and 6000 Hz at 20 dB.

Stuttering Diagnosis

We collected 750–1,000 syllable-spontaneous speech samples from each child during two play-based sessions, one with the child's primary caregiver and the other with the project speech-language pathologist (SLP). From these recordings, we calculated a weighted stuttering index (WSI) based on the frequency of part- and single-syllable whole-word repetitions, the number of iterations, and the presence and duration of sound prolongations or blocks per 100 syllables of spontaneous speech. A score of 4.0 or higher indicated the presence of childhood stuttering (Ambrose & Yairi, 1999). We also administered the Test of Childhood Stuttering (TOCS; Gillam, Logan, & Pearson, 2009), a norm-referenced assessment of stuttering severity. A score < 85 on this measure indicates stuttering. Finally, the parent and project SLP experienced in early childhood stuttering each rated the child as stuttering by assigning them a score of 2 or higher on an 8-point scale (0–1 =

normal, 2–3 = mild stuttering, 4–5 = moderate stuttering, and 6–7 = severe stuttering). The clinician's rating was based upon the type, duration, and frequency of disfluencies along with the presence and severity of secondary characteristics. Participant characteristics for each of the 22 CWS are provided in Appendix A. Three children received a WSI < 4.0; however, we retained these children in the study because they met the criteria for stuttering on the TOCS and per clinician and parent ratings.

Experimental Task

This experiment examined SNS arousal recorded during fluent and stuttered utterances produced during a picture description task. The children were asked to describe black-and-white-illustrated picture scenes (Webber, Webber, & Bristol, 1993). Prior to the start of data collection, one experimenter explained to the child that they would “see some pictures with different things happening in them.” Furthermore, they were told “There is no wrong answer; everyone sees something different in the pictures. We want to know what you see!” The experimenter then modeled what the child would do for two picture scenes and had them practice describing two different pictures to ensure that they understood the task and could describe the pictures using connected speech, rather than simply listing items they recognized in the pictures. This experimenter sat with the child and presented the picture scenes, while a second experimenter monitored the signals noting the approximate time intervals for fluent or stuttered utterances within the longer record. These intervals were used as a general guide for later off-line transcription and analysis. The second experimenter also noted the approximate time of significant movement artifact. Within a trial, each picture scene was separated by 5 s of rest. We collected each child's verbal responses and sympathetic recordings during two trials of five pictures for a minimum of 10 total picture scenes.

Data Acquisition

We used a Biopac MP150 data acquisition system (Biopac Systems, Inc.) running AcqKnowledge 4.4 acquisition software to collect SNS data. Children sat at a small desk while the experimenter affixed pregelled self-adhesive Ag/AgCl electrodes to the hypothenar eminence and thenar eminence of their nondominant hand. A Biopac GSR100C amplifier recorded skin conductance, reflecting eccrine sweat gland secretion (Boucsein, 2013), between these two electrodes at a 2.5-kHz sampling rate with an initial gain of 10 μ S/V and low-pass filtered at 1 Hz. BPV was recorded using a Biopac Pulse Plethysmograph amplifier (PPG100C) and transducer (TSD200) that were secured around the distal phalanx of the fourth finger of the participants' nondominant hand. The BPV signal was collected with a gain of 50 at a 2.5-kHz sampling rate.

We presented the picture scenes on a 25-in. monitor using Microsoft PowerPoint. The participants' utterances

were collected with a Shure SM90 tabletop condenser microphone at 10 kHz, and video recordings of the experimental session were made with a Logitech HD 720p webcam. Audio and video signals were synchronized with the autonomic recordings through the Biopac system. The video and audio recordings were used for off-line transcription and to ensure that utterances used in subsequent analyses did not contain movement artifact, extraneous comments, or redirection from the experimenter.

We segmented connected speech into utterances or verbal productions bounded by grammatical closure, terminal intonation contours, or long pauses following Systematic Analysis of Language Transcripts conventions (Miller & Iglesias, 2006). Given that SNS responses typically occur within 1–3 s of a stimulus, we selected fluent utterances that were not immediately adjacent (i.e., less than 3 s) to a stuttered utterance. One experimenter, a research assistant with more than 20 years of experience on the Purdue Stuttering Project, coded utterances from the picture description task as fluent or disfluent (stuttered) and tabulated the number of syllables for each response. An utterance was considered stuttered if it contained an SLD: part-word or single-syllable whole-word repetition, sound prolongation, broken word, or block. Extra sound, syllable, or single-syllable whole-word repetitions were not included in the syllable count for the stuttered utterances. Given that our picture description task elicited spontaneous, connected speech production, all participants produced typical disfluencies such as filled pauses and multisyllabic word repetitions. Typical disfluencies occurring during stuttered and fluent (nonstuttered) utterances were also omitted from the syllable count. The first author re-analyzed 20% of the data to establish interrater reliability for utterance coding. The average intraclass correlation was .95, with a 95% confidence interval from .91 to .98 indicating excellent reliability.

Data Processing and Analysis

Raw SNS data were processed using a custom program (Matlab Version 2018b). The skin conductance signal was down-sampled to 250 Hz, and phasic SCRs were derived from the tonic skin conductance signal through high-pass filtering (Fpass 0.07 Hz/Apass 0.5 dB). The BPV signal was also down-sampled to 250 Hz and digitally bandpass filtered between 0.5 and 3 Hz with a 100-order finite impulse response filter (Arnold, MacPherson, & Smith, 2014). A graphic user interface presented the acoustic, skin conductance, and BPV signals in a waterfall display. The video record, synchronized to the autonomic recordings, was displayed on an adjacent monitor. The experimenter extracted fluent or stuttered utterances from the longer, continuous recordings by indicating onset (i.e., start of speech) and offset (end of speech using cues described above) points in the acoustic record with a cursor (see Figure 1). However, we adjusted onset and offset indices to ensure that segmented utterances did not include motion artifact. For example, if the child said, “I see a tiger in a cage with big teeth and (movement occurs) he is eating

Figure 1. Sympathetic nervous system recordings from a child who stutters during the picture description task. The waveforms represent, from top to bottom, the acoustic signal, blood pulse volume (BPV) signal, and phasic skin conductance response (SCR). Utterances were extracted from the continuous recording by identifying speech onsets and offsets within the acoustic record.

meat,” the experimenter would place the cursor at the onset of speech, in this example, “I,” and place the offset after “and” to avoid the motion artifact occurring in the later part of the utterance. The accompanying SCR and BPV measures were automatically extracted using the onset/offset indices of the segmented utterance during this procedure. We analyzed all usable fluent and disfluent utterances (up to 10), which yielded six to 10 fluent and six to 10 stuttered utterances, each lasting, on average, 8 s for each child (Arnold et al., 2014).

Experimental Variables

Skin Conductance Measures

For selected fluent and stuttered utterances, we calculated two indices capturing phasic changes in skin conductance (Boucsein, 2013): (a) SCR amplitude,¹ an amplitude measure expressed in microSiemens (μS) of the phasic SCRs that occurred during fluent or stuttered speech utterances. SCR amplitude is derived by subtracting the minimum SCR value from the maximum SCR value associated with the largest SCR peak (if any) for each extracted production. An average was then taken across all stuttered or fluent utterances. Amplitude of SCRs indicates level of SNS arousal. (b) SCR frequency or the number of SCRs $\geq 0.05 \mu\text{S}$ was tabulated for each utterance and an average was taken across all fluent or stuttered utterances. SCRs occurring at a higher frequency are also indicative

¹We refer to this measure as amplitude for familiarity; however, we computed the SCR magnitude, the more commonly used index that calculates a mean across all responses. This allows for an approximately equal number of trials contributing to each participant’s overall average (Boucsein et al., 2012).

of SNS arousal. It is important to note that SCRs are not elicited in all trials (Andreassi, 2000). However, we included utterances with and without clear SCRs in the calculation of the mean. In these cases, the SCR frequency and SCR amplitude would be ~ 0 . We computed paired-samples *t* tests to compare phasic skin conductance activity occurring during children’s fluent versus stuttered utterances. Bonferroni-corrected alpha level was set at $p < .025$, accounting for the two measures of skin conductance. Effect sizes were also calculated for each *t* test using Cohen’s *d*. Standard interpretation of this index—*d* of 0.20 = small effect, *d* of 0.50 = moderate effect, and *d* of 0.80 = large effect—was applied.

Blood Pulse Measures

We derived two measures of BPV (Arnold et al., 2014): (a) average BPV amplitude, computed by taking the trough-to-peak amplitude (in volts) of pulse cycles from each fluent or stuttered production using an automatic peak-selecting algorithm, and (b) average pulse rate (in pulses per minute) calculated for stuttered and fluent utterances. As with skin conductance analysis, we used paired-samples *t* tests ($p < .025$) and Cohen’s *d* effect sizes to compare BPV and pulse rate measures collected during fluent versus stuttered utterances.

Behavioral Measures

In addition to using the TOCS and WSI to capture stuttering severity, we administered the Communication Attitude Test for Preschool and Kindergarten Children Who Stutter (KiddyCAT; Vanryckeghem & Brutten, 2006). Children answered 12 yes/no questions that assessed overall attitude about their speech. Children who receive higher scores on this measure are considered to have more negative

feelings about their speaking abilities. We computed Pearson correlations to assess the relationship among SNS arousal, stuttering severity (TOCS and WSI scores), and communication attitude (KiddyCAT scores). For SNS variables (SCR amplitude/SCR frequency and BPV amplitude/pulse rate), we computed the percent change by subtracting a participant's fluent value from their disfluent value and expressing this difference as a percentage of their fluent value to use in the correlations.

Results

Behavioral Measures

The mean score on the KiddyCAT was 3.14 ($SD = 3.11$, range: 0–10; see Appendix A), which is below the mean reported for 3- to 6-year-old CWS but higher than the mean for 3- to 6-year-old CWNS on this measure (e.g., CWS $M = 4.36$ and CWNS $M = 1.79$ in Vanryckeghem & Brutten, 2006). Approximately one third of CWS scored higher than the reported mean (5–10 out of 12 possible), while, the rest of the children, ~67% scored 3 or below. Approximately 45% of children answered “yes” to the item “Do you think people need to help you talk?” Approximately 38% of CWS answered “no” to the item “Do you think your words come out easily?”, 33% of CWS answered “yes” to “Do words sometimes get stuck in your mouth?”, and 33% answered “no” to “Do you think you talk right?”

There was no difference in the average number of fluent ($M = 8.9$, $SD = 1.8$) and stuttered ($M = 9.1$, $SD = 1.8$) utterances available for analysis, $t(21) = -0.67$, $p = .51$. The children's fluent utterances ($M = 8.3$, $SD = 0.8$) were longer, on average, by 0.80 s than their stuttered utterances ($M = 7.5$, $SD = 1.1$), $t(21) = 2.99$, $p < .01$. Fluent utterances ($M = 11.8$, $SD = 2.7$) contained, on average, 1.5 more syllables than stuttered utterances ($M = 10.3$, $SD = 2.3$), $t(21) = 3.62$, $p < .01$. Utterances that are slightly longer in duration and number of syllables may incur higher sympathetic arousal. Since the finding was noted for fluent utterances, however, it does not present a confound to our hypothesis that stuttered utterances would be associated with higher arousal. Finally, we did not detect a difference in the number of typical speech disfluencies occurring during fluent ($M = 4.0$, $SD = 3.9$) and stuttered ($M = 5.1$, $SD = 3.4$) utterances, $t(21) = -1.35$, $p = .19$. Stuttered utterances contained a total of 343 SLDs. Single-syllable whole-word repetitions and part-word repetitions made up the majority, or 52% and 31%, respectively, of the total SLDs. Prolongations or broken words made up 16% of the total SLDs, while the percentage of blocks was < 1%. Appendix B provides additional details about the stuttered and fluent utterances.

Psychophysiological Measures

Skin Conductance

Overall, we detected significantly higher SNS arousal, indexed by SCR measures, during stuttered speech

production compared to fluent speech production. As shown in Figure 2a, mean SCR amplitude was significantly higher for stuttered speech compared to fluent speech, $t(21) = 4.65$, $p < .01$, $d = 0.99$, representing a large effect. SCR frequency also increased during stuttered speech compared to fluent speech (see Figure 2b), $t(21) = 3.01$, $p < .01$, $d = 0.64$, representing a moderate effect. In Figures 2c and 2d, we computed difference scores in mean SCR amplitude and SCR frequency, respectively, from the participants' stuttered and fluent utterances by subtracting their average fluent value from their average stuttered value for these variables. With a few exceptions, most of the children demonstrated higher SCR amplitudes and increased SCR frequencies while stuttering compared to when they were speaking fluently.

Blood Pulse Measures

We recorded lower mean BPV amplitude during stuttered utterances compared to fluent utterances, reflecting a greater degree of vasoconstriction during stuttering, $t(21) = -2.49$, $p = .02$, $d = -0.53$, a moderate difference in these means (see Figure 3a). A majority of our participants exhibited negative mean difference scores (see Figure 3c) as BPV amplitude decreased with increased SNS arousal. The overall mean increase in pulse rate during stuttered speech missed statistical significance (see Figure 3b), $t(21) = 1.95$, $p = .07$, $d = 0.42$. A clear pattern in the individual responses was not as apparent for pulse rate (see Figure 3d). Although there were several children with higher pulse rates during stuttered speech, other children showed smaller changes in pulse rates that went in either direction (i.e., faster or slower).

Correlations Among Autonomic and Behavioral Measures

We did not find significant relationships among indices of SNS arousal and stuttering severity (WSI scores) or communication attitude (KiddyCAT scores). Pearson correlations between WSI scores and SNS variables were not statistically significant ($ps = .38-.89$). As two thirds of the children's KiddyCAT scores fell below the mean reported by Vanryckeghem and Brutten (2006), we did not find significant correlations between KiddyCAT scores and SNS measures ($ps = .16-.89$). Finally, the correlation between stuttering severity indexed by the WSI and the KiddyCAT was also not significant, $p = .16$.

Preliminary Post Hoc Language Analysis

Stuttering is more likely to occur during the production of longer or more complex utterances (Bloodstein & Ratner, 2008), and ANS responses are sensitive to increased task demands and cognitive effort (Iani et al., 2004; MacKersie & Calderon-Moultrie, 2016; Tulen et al., 1989). We wished to examine further the finding of increased SNS activity during stuttered utterances by assessing whether the fluent/stuttered utterances differed on standard expressive language measures. We conducted an exploratory post hoc language analysis of children's utterances using Computerized Language Analysis (MacWhinney, 2000) and

Figure 2. (a) Mean skin conductance response (SCR) amplitude and (b) SCR frequency across stuttered and fluent responses. Difference scores were computed for each participant for (c) SCR amplitude and (d) SCR frequency. Amp = amplitude; PK = peak; CWS = children who stutter.

examined mean length of utterance in morphemes, number of different words, vocabulary diversity, and Brown's morphemes (present participle, regular past tense, copula, determiner/article, uncontracted auxiliary). We found no significant differences between the two classes of utterances on these measures ($ps = .21-.91$; see full results in Appendix B). We had far fewer fluent and stuttered utterances from each participant than the minimum recommended number of utterances to conduct a valid language sample analysis (~50). As a result, a number of standard analyses returned "NA" values. These findings should therefore be interpreted as preliminary.

Discussion

This is the first study to examine SNS activity associated with fluent and stuttered utterances in preschool CWS. We assessed phasic indices of SNS arousal during connected speech produced during a picture description task and compared psychophysiological responses in the context of fluent and stuttered utterances. Skin conductance and BPV measures contributed to an overall picture of higher SNS arousal during stuttered speech compared to fluent speech. Specifically, we detected larger amplitude SCRs occurring at a greater frequency and decreased BPV

Figure 3. (a) Mean blood pulse volume (BPV) amplitude and (b) mean pulse rate across stuttered and fluent utterances. Difference scores were computed for each participant for (c) BPV amplitude and (d) pulse rate. Amp = amplitude; PR = pulse rate; CWS = children who stutter.

amplitudes during children's stuttered utterances compared to their fluent utterances. As Figures 2c, 2d, and 3c illustrate, the pattern of higher arousal associated with stuttered speech was seen across the majority of participants, albeit to different degrees. Pulse rate differences between fluent and stuttered utterances did not reach statistical significance. Despite the observed differences in SNS arousal associated with the occurrence of stuttering, we did not find significant relationships between SNS indices and stuttering severity, corroborating findings from Choi et al. (2016), nor did we find significant relationships between SNS indices and communication attitudes (KiddyCAT scores). Finally, our exploratory language analysis of stuttered and fluent

utterances yielded no discernable differences between the two classes of utterances on any expressive language measure (see Appendix B).

These findings raise questions about the potential association between SNS arousal and speech fluency, including underlying mechanisms involving the interaction between speech motor and ANS activity. Interpretation of the putative relationship between high SNS arousal and stuttered speech is constrained by several unknowns. First, the directional relationship between SNS arousal and disfluency, for example, if increases in SNS arousal contributed to or resulted from stuttering, is difficult to determine. Second, the low temporal resolution of autonomic indices

restricts understanding of the task-dependent motoric, cognitive, and emotional variables that may contribute to heightened SNS arousal. Unlike previous investigations examining ANS activity associated with different emotional states in preschool CWS and CWNS (e.g., Jones, Buhr, et al., 2014), our experiment did not attempt to induce different emotional states; rather, we observed SNS activity during overt speech production. Nevertheless, peripheral physiological responses must be interpreted according to the situational context (Mendes, 2016). In this case, children had to generate novel language and speak aloud in front of unfamiliar adults. When the expression of their thoughts was interrupted by involuntary breakdowns in the forward flow of their speech, it is reasonable to interpret increases in sympathetic arousal as reflective of negative emotion rather than joy, for example. Moreover, the indicators of awareness that Boey et al. (2009) documented during moments of stuttering were predominantly negative, for example, verbal remarks and nonverbal displays conveying sadness and frustration.

Potential Mechanisms Underlying Increased Sympathetic Arousal During Stuttered Speech

Given that we observed differences in phasic SNS arousal during the fluent and stuttered speech of the same CWS, sympathetic autonomic arousal may be a salient factor in their stuttering. Although we cannot discern precisely what precipitated increased SNS arousal during stuttering, it is possible that an increase in SNS activity during speech production contributed to breakdowns in the vulnerable speech sensorimotor systems of CWS. Corroborating our finding of higher arousal during stuttered utterances in preschool CWS, Weber and Smith (1990) also found higher SNS arousal during stuttered speech compared with fluent speech in AWS—evidence that task-specific arousal is associated with stuttering in some CWS and AWS.

Pertinent questions emerge regarding the role that SNS arousal plays in disfluent speech, specifically whether shifts in SNS arousal contributed to the likelihood of speech breakdowns or whether phasic increases in SNS activity were a response to the disfluency. Theoretical accounts of the well-documented anticipation effect in stuttering propose that anticipation, fear, and avoidance of a stuttering moment, coupled with an overly sensitive speech monitoring system, are significant factors in stuttering in older children and adults (Bloodstein & Ratner, 2008; Garcia-Barrera & Davidow, 2015; W. Johnson, 1938; Vasić & Wijnen, 2005). Moments of stuttering have traditionally been associated with negative emotional or psychological states, such as a “loss of control” (Perkins, 1990) or “anticipatory struggle” (Bloodstein & Bernstein Ratner, 2008). Other theories maintain that negative emotions elicited by stuttering, along with a reduced ability to regulate these emotions, may increase one’s vulnerability to stuttering (Brutten & Shoemaker, 1967; Walden et al., 2012).

In Walsh et al. (2019), average tonic SCL measures were significantly higher in preschool CWS compared to CWNS during speech and nonspeech task performance (but not at baseline). However, phasic measures of SNS arousal recorded from CWS and CWNS during fluent speech did not differ. Therefore, findings of increased SNS arousal in preschool CWS during stuttered speech compared to fluent speech may reflect different emotional and/or speech regulatory mechanisms specifically related to stuttering behaviors. For example, the increase in SNS arousal may be a response to the involuntary disruptions that disrupt a child’s speech. Self-awareness of speech struggles and negative feelings toward communication are emerging in preschool CWS (Boey et al., 2009; Langevin, Packman, & Onslow, 2010; Vanryckeghem & Brutten, 1997). Studies showed that stuttering moments are associated with negative emotional reactions in preschool CWS (Jones, Buhr, et al., 2014), and more frequent stuttering is associated with greater negative emotion (Walden et al., 2012). We found that only one third of preschool CWS had scores indicating negative communication attitudes, although many children’s responses conveyed the sentiment that speech was difficult. It is also important to note that we did not include more nuanced indices signifying awareness or the expression of negative feelings such as the nonverbal and verbal indicators that Boey et al. (2009) recorded. It is possible that increased SNS arousal during stuttered utterances reflects implicit or explicit awareness of speech breakdowns, increased effort to maintain fluent speech, or, in some children, negative emotion toward involuntary speech disruptions.

Regardless of causal mechanisms of increased sympathetic arousal, it is clear that the ANS acts as an intermediary between brain and body: Top-down processes influence somatic physiological states, while even small shifts in physiological arousal can, in turn, influence our thoughts and behaviors (e.g., Critchley, 2009). This overarching framework is consistent with multifactorial (A. Smith & Weber, 2017), dual-diathesis (Walden et al., 2012), and demands-and-capacities (Starkweather & Gottwald, 1990) views of the potential role of emotional factors in stuttering. Increased SNS outflow may have bidirectional influences on speech production in preschool CWS. Against elevated tonic levels of SNS arousal noted in our companion study (Walsh et al., 2019), phasic increases in arousal may impart a greater load for emotional and speech regulatory mechanisms and impoverishing resources for fluent speech production. The misallocation of neural resources is also central to classic and contemporary theories of stuttering (Adams, 1990; Andrews & Harris, 1964; Arenas, 2017; Bosshardt, 2006; A. Smith & Weber, 2017; Starkweather & Gottwald, 1990).

Effects of SNS Arousal on Speech and Other Motor Behaviors

It is reasonable to speculate about the potential role of SNS arousal on speech motor control in stuttering from

a conceptual standpoint, yet the networks interconnecting the ANS and higher cortical regions that mitigate motor, cognitive, and emotional functions are complex and not fully understood (Boucsein, 2013; Cardinali, 2018; Jänig, 2006). Investigations of overt motor performance under different task conditions confirm that higher levels of sympathetic arousal negatively impact motor control and coordination for high-precision tasks such as biathlon or piano playing as well as for simple grip tasks in adult participants (Noteboom, Fleshner, & Enoka, 2001; Vickers & Williams, 2007; Yoshie, Kudo, & Ohtsuki, 2009; Yoshie, Nagai, Critchley, & Harrison, 2016). The effects of sympathetic arousal on motor performance extend to children. Spatiotemporal analysis of children's stepping revealed less efficient and smooth movement trajectories while children were being evaluated by an observer compared to unobserved conditions (Beuter & Duda, 1985; Beuter, Duda, & Widule, 1989). These findings confirm that, during voluntary movement, there is temporal coupling between central motor commands to muscles and SNS activation to skin.

While these studies have documented the clear effects of increased sympathetic arousal on other motor behaviors, investigations examining the influence of autonomic arousal on speech motor control are scant. Speech production is associated with relatively high levels of autonomic arousal in typical speakers and in people who stutter (Arnold et al., 2014; Mackersie & Calderon-Moultrie, 2016; Peters & Hulstijn, 1984; Weber & Smith, 1990). Several accounts suggest a relationship between arousal and speech performance in typical speakers. Kleinow and Smith (2006) assessed the spatiotemporal coupling of upper lip, lower lip, and jaw movements in typically speaking adults and school-age children under lower and higher arousal speaking conditions induced by a Stroop speaking condition. They found that the Stroop speaking condition was associated with ANS arousal, evidenced by increases in skin conductance and heart rate and reduced BPV, and decreased coordination of articulatory coupling during sentence production. In a recent study of the effects of autonomic arousal on voice production in 16 healthy young adults, MacPherson, Abur, and Stepp (2017) found a significant relationship between skin conductance measures and acoustic phonatory parameters under increased cognitive load (also elicited by a Stroop task). Furthermore, Tolkmitt and Scherer (1986) noted that the effects of stress on phonatory and articulatory measures were significantly greater in participants who self-reported being more susceptible to stress. Albeit conducted with typically fluent speakers, these studies offer collective support for the hypothesis initially proposed by Zimmermann (1980) that aberrant sympathetic activity related to increased arousal could negatively affect speech production processes.

Limitations and Future Research Directions

Although we cannot rule out the possibility that increased SNS arousal recorded during stuttered speech was an epiphenomenon of stuttering, we adopted measures

against this possibility. For instance, body movement may produce an SNS response (Boucsein, 2013). We reviewed the accompanying video for each fluent and stuttered utterance from the picture description task to ensure that tokens used in the analysis were free from movement artifact. Anomalous respiratory patterns, for example, breath holds and deep inhalation, can also elicit SNS responses (Hygge & Hugdahl, 1985). We did not record respiratory kinematics concurrent with SNS measures; however, our analysis of SLD type revealed that silent blocks—abrupt stoppages of airflow that disrupt breathing patterns—made up < 1% of the total SLDs.

There is considerable within- and between-individual variability in autonomic measurements (Dawson et al., 2007). Therefore, experimenters have previously recorded task-related changes in SNS arousal relative to a baseline collected at the onset of each experimental task to account for changes in arousal levels over the course of the experiment. Arnold et al. (2014) adopted this strategy for their study of SNS arousal in school-age children and adults collecting a 30-s baseline before each task. They also selected three intervals to measure SNS arousal related to portions of the task: 5 s prior to onset, 5 s during the task, and 5 s immediately after the task. SNS arousal during these three intervals was then expressed as a percentage change relative to the pretask baseline. Their findings, however, were difficult to interpret given that, in some cases, arousal was higher during baseline than the task resulting in negative percent change. It is possible that pretask baselines were not long enough to allow SNS arousal to return to baseline levels. Moreover, because the onset of a response sometimes began during one interval but peaked in another, Arnold et al. adopted a rule regarding which interval to “count” the response. Given these methodological limitations, we adopted a different approach in the current study to accommodate data collection with preschoolers. We examined within-participant differences in phasic SNS arousal during fluent/stuttered utterances produced during the same task. Thus, children served as their own control. We elected not to divide task intervals into pre, during, and post phases; rather, we examined responses that occurred during the selected onset/offsets for each utterance. The average onset latency of BPV constriction is approximately 1 s and, for SCRs, between 1 and 3 s (Andreassi, 2000; Dawson et al., 2007; Lim, Seto-Poon, Clouston, & Morris, 2003). The further away from the stimuli (i.e., fluent/stuttered utterance), the less confident we could be regarding the timing of an SNS response and the greater the opportunity for contamination by motion or other artifacts.

There is clearly much to learn about the relationship among increased SNS arousal, stuttering behaviors, and speech motor development in CWS. We pose several questions here to motivate future research: (a) Do psychophysiological markers of increased autonomic arousal in young CWS confer a greater risk for persistent stuttering? A recent longitudinal study offers preliminary evidence to support this. CWS who would eventually persist demonstrated significantly higher SNS arousal during a stress-inducing

naming task compared to CWS who would eventually recover (Zengin-Bolat kale et al., 2018). (b) What influence does increased SNS arousal have on articulatory, laryngeal, or respiratory physiology in people who stutter? (c) What neural processes (e.g., emotional, cognitive, linguistic, motoric) elicit the increased autonomic arousal associated with stuttering? (d) Does increased sympathetic arousal during stuttered utterances in some CWS portend future negative communication attitudes and anxiety?

Conclusions

We recorded larger amplitude SCRs, a greater frequency of SCRs, and decreased BPV amplitudes during children's stuttered compared to fluent utterances. Our findings collectively support the conclusion that some preschool CWS exhibit higher levels of sympathetic arousal during stuttered speech compared to when they are speaking fluently. However, we did not find a clear relationship among indices of SNS arousal and stuttering severity or communication attitudes per our self-report measure (Kiddy-CAT). These results contribute to the formation of a data-driven framework that integrates influences of SNS arousal on speech production in preschool CWS.

Although we noted increased SNS arousal associated with stuttered speech in preschool CWS, these data represent a single developmental time point whose awareness of stuttering is emerging. Children who persist in stuttering will undoubtedly encounter negative communication experiences—adverse reactions, teasing, bullying, and repeatedly failed attempts to produce fluent speech when it is most desired (Blood & Blood, 2004; Blood, Blood, Maloney, Meyer, & Qualls, 2007; Guttormsen et al., 2015; K. A. Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014; Vanryckeghem & Brutten, 1997; Vanryckeghem, Hylebos, Brutten, & Peleman, 2001). The coupling of negative reactions, aberrant sympathetic activity, and speech breakdowns could contribute to a chronic disorder and higher levels of communication anxiety. This speculation awaits empirical support through longitudinal studies with CWS.

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Appendix A

Participant Characteristics

Participant	Sex	Age	Reported age at onset (months)	Time since onset (months)	History of therapy for stuttering	WSI	TOCS	Clinician rating ^a	KiddyCAT
1	M	71	36	35	N	2.72	80	Mild	0
2	M	58	55	3	N	5.75	71	Mild to moderate	1 ^b
3	M	48	30	18	N	23.42	58	Moderate	5
4	F	52	36	16	N	16.40	58	Moderate	8
5	M	51	41	10	Y	9.36	58	Moderate	0
6	M	62	42	20	N	3.73	62	Mild	0
7	M	50	21	29	N	10.25	58	Mild to moderate	7
8	M	46	24	22	N	6.40	65	Mild to moderate	0
9	M	57	36	21	N	12.07	58	Moderate	0
10	M	59	36	23	N	14.11	58	Moderate	1
11	M	53	28	25	N	9.73	58	Moderate	3
12	M	50	36	14	N	12.93	58	Moderate	1
13	M	52	46	6	N	13.41	58	Moderate	3
14	M	47	32	15	N	13.77	58	N/A	3
15	F	59	42	17	Y	5.62	58	Moderate	0
16	M	48	34	14	N	32.61	58	Moderate	2
17	F	61	32	29	N	3.76	58	Moderate	7
18	M	50	42	8	N	10.06	58	Moderate	6
19	M	48	42	6	N	9.48	58	Mild	7
20	M	50	36	14	Y	11.07	58	Mild to moderate	3
21	M	51	30	21	N	22.21	58	Moderate	10
22	M	50	24	26	N	13.19	58	Mild to moderate	2

Note. WSI = weighted stuttering index; TOCS = Test of Childhood Stuttering; KiddyCAT = Communication Attitude Test for Preschool and Kindergarten Children Who Stutter; M = male; N = no; F = female; Y = yes; N/A = data not available.

^a0–1 = *normal*, 2–3 = *mild*, 4–5 = *moderate*, and 6–7 = *severe* (midpoints may be selected). Clinician rating was based upon the type, duration, and frequency of disfluencies along with the presence and severity of secondary characteristics. ^bOnly responded to a total of four out of 12 items per parent request.

Appendix B

Utterance Information

Table B1. Utterance information.

Variable	Stuttered	Fluent
Total number of utterances	203	196
Total number of utterances with SCRs ($\geq 0.05 \mu\text{S}$)	147 (72.4%)	122 (62.2%)
Average number of typical disfluencies per utterance	0.41 (0.39)	0.57 (0.33)
Average number of SLDs per utterance	1.87 (0.47)	N/A

Note. SCRs = skin conductance responses; SLDs = stuttering-like disfluencies; N/A = not applicable.

Table B2. Results of preliminary comparison of stuttered and fluent utterances on standard expressive language measures.

Variable	Stutt	Fluent	Mean difference (SD)	95% CI		<i>t</i>	<i>df</i>	Sig. (two-tailed)
	<i>M</i> (SD)			Lower	Upper			
MLU in morphemes	3.54 (1.35)	3.46 (1.34)	0.07 (1.55)	-0.61	0.76	0.22	21	.83
Number of different words	50.09 (14.13)	49.64 (13.67)	0.46 (18.06)	-7.55	8.46	0.12	21	.91
Vocabulary diversity	24.74 (11.46)	26.95 (12.46)	-2.21 (18.16)	-10.26	5.84	-0.57	21	.57
Brown's morphemes stuttering								
Present participle <i>-ing</i>	3.50 (2.40)	3.59 (2.95)	-0.09 (4.04)	-1.88	1.70	-0.11	21	.92
Regular past tense	1.23 (1.15)	1.50 (1.47)	-0.27 (1.88)	-1.10	0.56	-0.68	21	.50
Copula	1.05 (1.17)	1.64 (1.56)	-0.59 (2.15)	-1.55	0.36	-1.29	21	.21
Determiner-article	10.82 (6.02)	9.95 (5.79)	0.86 (9.47)	-3.34	5.06	0.43	21	.67
Uncontracted auxiliary	3.36 (3.76)	2.86 (2.82)	0.50 (4.37)	-1.44	2.44	0.54	21	.60

Note. CI = confidence interval; Sig. = significance; MLU = mean length of utterance; Stutt = stuttering; Flu = fluent.