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# SYMPATHETIC AROUSAL AS A MARKER OF CHRONICITY IN CHILDHOOD STUTTERING

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## Abstract

**Purpose—**This study investigated whether sympathetic activity during a stressful speaking task was an early marker for stuttering chronicity.

**Method—**Participants were 9 children with persisting stuttering, 23 children who recovered, and 17 children who do not stutter. Participants performed a stress-inducing picture-naming task and skin conductance was measured across 3 time points.

**Results—**Findings indicated that at the initial time point, children with persisting stuttering exhibited higher sympathetic arousal during the stressful speaking task than children whose stuttering recovered.

**Conclusions—**Findings are taken to suggest that sympathetic activity may be an early marker of heightened risk for chronic stuttering.

## Introduction

Developmental stuttering is characterized by a disruption in the forward flow of speech resulting in speech disfluencies such as repetitions of sounds and syllables and sound prolongations (for further data-based descriptions of characteristics of childhood stuttering, see Tumanova, Conture, Lambert, & Walden, 2014). The disorder typically begins between 2-5 years of age, with an incidence of approximately 5% of preschool-age children affected; however, 75-80% of children who begin to stutter eventually drop below the threshold of the diagnostic criteria (for review, see Yairi & Ambrose, 2013). For the remaining 1% of children that continue to stutter there is significant potential for deleterious impact on social-emotional (McAllister, 2016; Van Borsel, Brepoels, & De Coene, 2011), educational (O'Brian, Jones, Packman, Menzies, & Onslow, 2011), and vocational (Klein & Hood, 2004) aspects of life. Therefore, there is a critical need to improve diagnostic capabilities to identify those at high risk to continue to stutter into school-age years, adolescence, and adulthood to enhance clinical outcomes.

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## Persistence of Childhood Stuttering

To date, numerous factors have been associated with increased risk for persistent stuttering. For example, greater time since onset (TSO; i.e., the length of time a child has been stuttering) is significantly associated with increased risk for persistence (Anderson & Felsenfeld, 2003; Pellowski & Conture, 2002; Yairi, Ambrose, Paden, & Throneburg, 1996). Males have been shown to be at increased risk to persist than females (Ambrose, Cox, & Yairi, 1997), with children whose caregiver(s) report a family history of chronic stuttering being more likely to persist than those with a family history of recovered stuttering or no family history of stuttering (Ambrose et al., 1997).

Findings from longitudinal investigations support theories that speech-motor, linguistic, and emotional or temperamental factors (e.g., Adams, 1990; Conture & Walden, 2012; Smith & Weber, 2016) are associated with developmental trajectories of persistent stuttering. For instance, children who stutter and persist differ from those who recover and those who do not stutter with regard to: (1) speech-motor coordination (e.g., Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Usler, Smith, & Weber, 2017; Yaruss & Conture, 1993), (2) speech articulation and phonology (e.g., Paden, Yairi, & Ambrose, 1999; Spencer & Weber-Fox, 2014), (3) linguistic processes (e.g., Ambrose et al., 2015; Mohan & Weber, 2015; Usler & Weber-Fox, 2015; Yairi et al., 1996), (4) vocabulary abilities (e.g., Ambrose et al., 2015; cf. Spencer & Weber-Fox, 2014), and (5) temperament (e.g., Ambrose et al., 2015). Further, differences in white matter (Chang, Zhu, Choo, & Angstadt, 2015) and intra- and inter-network connectivity (Chang et al., in press) that support speech-motor and task-related attentional processes have been reported for children who persist compared to those who recover and those who do not stutter. Despite these recent advances, there is still a need for the continued development of markers of risk for persistent stuttering. The present study focuses on emotion—a theoretically motivated (e.g., Conture & Walden, 2012) and empirically substantiated contributor to early childhood stuttering (e.g., Eggers, De Nil, & Van den Bergh, 2010) that has received little longitudinal investigation as a potential marker of risk for chronic stuttering.

## Emotion and Childhood Stuttering

A recent theoretical account of childhood stuttering suggests that emotional processes may also be associated with the onset and the development of childhood stuttering (Conture & Walden, 2012). Furthermore, findings from numerous empirical studies conducted over the past decade lend support to the possible association between emotional processes and childhood stuttering (e.g., Anderson, Pellowski, Conture, & Kelly, 2003; Arnold, Conture, Key, & Walden, 2011; Choi, Conture, Walden, Jones, & Kim, 2016; Choi, Conture, Walden, Lambert, & Tumanova, 2013; Clark, Conture, Walden, & Lambert, 2015; Eggers, De Nil, & Van den Bergh, 2010, 2012, 2013; Felsenfeld, van Beijsterveldt, & Boomsma, 2010; Johnson, Walden, Conture, & Karrass, 2010; Jones, Buhr, et al., 2014; Jones, Conture, & Walden, 2014; Jones, Walden, Conture, Erdemir, Lambert & Porges, 2017; Karrass et al., 2006; Ntourou, Conture, & Walden, 2013; Schwenk, Conture, & Walden, 2007; Walden et al., 2012; Zengin-Bolatkale, Conture, & Walden, 2015).

Using various methodological approaches (i.e., caregiver report, behavioral observation, and psychophysiology), results of the above studies indicated that compared to children who do not stutter, the following have been shown to be characteristics of children who stutter as a group: (1) higher emotional reactivity to emotional stimuli (Jones, Buhr, et al., 2014; Jones, Conture, et al., 2014; Karrass et al., 2006; Ntourou et al., 2013; Zengin-Bolatkale et al., 2015), (2) lower efficiency in regulating emotion and attention (Eggers et al., 2012; Felsenfeld et al., 2010; Jones, Buhr, et al., 2014; Karrass et al., 2006), (3) more negative affect during experimental conditions (Jones, Conture, et al., 2014; Ntourou et al., 2013), and (4) less adaptability (Anderson et al., 2003; Schwenk et al., 2007). Further, increased stuttering frequency and severity have been associated with increased emotional reactivity and decreased regulation for children who stutter (Arnold et al., 2011; Choi et al., 2016, 2013; Jones et al., 2017; Kraft, Ambrose, & Chon, 2014; Walden et al., 2012).

Such empirical findings regarding emotional processes have been particularly valuable due to their focus on stuttering in early childhood, close to the onset and prior to children developing established emotional reactions resulting from experience with stuttering. However, although these studies have established that emotion and temperament are associated with childhood stuttering, they generally have not addressed whether these characteristics and processes are associated with developmental trajectories of recovered versus chronic stuttering. One exception are findings of an empirical study by Ambrose et al. (2015) indicating that children who stutter and exhibit persistent stuttering were rated by their caregivers as exhibiting greater negative affectivity than children who recovered and children who do not stutter. Therefore, emerging evidence suggests that emotional processes may be associated with persistent childhood stuttering. To further explore this possibility, the present study assessed whether emotional reactivity, indexed physiologically by sympathetic nervous system activity (i.e., skin conductance level), is a marker of increased risk for stuttering persistence.

## Sympathetic Nervous System Activity and Speech Production

The sympathetic nervous system—a component of the autonomic nervous system—facilitates adaptive responses of the endocrine, immune, sensory-motor and cognitive systems in response to physical or psychological challenges (Dawson, Schell, & Filion, 2007; Porges, 2007). One measure of sympathetic nervous system activity—electrodermal activity—has been documented to be a reliable index of arousal in response to emotional stimuli (e.g., Sequeira, Hot, Silvert, & Delplanque, 2009). Specifically, electrodermal activity, as indexed physiologically by changes in skin conductance level is associated with changes in sweat via the eccrine glands, which is exclusively regulated by the sympathetic branch of the autonomic nervous system (Boucsein et al., 2012). Thus, measuring skin conductance appears to provide a reliable index of emotional reactivity that can be used in a variety of experimental conditions and for this reason has been widely used in the empirical study of psychological processes (e.g., El-Sheikh, Keller & Erath, 2007; Figner & Murphy, 2011; McManis, Bradley, Berg, Cuthbert & Lang, 2001).

Speech-language planning and production, a process importantly involved with speech (dis)fluency, is a complex process (for an example of a theory that underscores the beadth and depth of speech-language planning and production, see Levelt, Roelofs, & Meyer, 1999). Such planning and production requires the coordination of neural systems involved with linguistic encoding, motor control, cognition, and emotion. Accordingly, sympathetic nervous system activity has been empirically studied during speech-related tasks in children (Arnold, MacPherson, & Smith, 2014; Zengin-Bolatkale et al., 2015) and adults (Peters & Hulstijn, 1984; Weber & Smith, 1990). Further, higher levels of sympathetic arousal (as indexed by skin conductance levels) have been found to impact children and adult's speech motor coordination (Kleinow & Smith, 2006). Specific to speech disfluencies, Weber and Smith (1990) reported that higher levels of skin conductance were associated with increased occurrence and severity of speech disfluencies in adults who stutter. Thus, these empirical studies provide support for the notion that sympathetic nervous system activity is associated with speech-language planning and production as well as breakdowns in these processes (e.g., stuttering).

## Childhood Stuttering and Skin Conductance

To date, the present authors are aware of two empirical studies that have empirically investigated the association between skin conductance level (SCL, as an index of emotional reactivity) and stuttering among young children in the age range close to stuttering onset and when trajectories of persistence and recovery are yet to be determined. Jones, Buhr, et al. (2014) reported that young children who stutter, compared to those who do not, exhibited increased skin conductance during a speaking task that followed positive emotional arousal (compared to neutral and negative arousal). Similarly, Zengin-Bolatkale et al. (2015) reported that 3-year-old children who stutter had greater SCL than 3-year-old children who do not stutter during a stressful picture-naming task, however, there were no differences between 4- and 5-year-old children who do and do not stutter. Based on these findings, there appear to be salient differences in skin conductance during speech between children who do and do not stutter, however, these associations are nuanced and may be impacted by various other factors (e.g., chronological age, emotion condition). Further, it is possible that these emotionally reactive responses during speech may be associated with increased risk for chronic stuttering. For example, increased physiological reactivity concurrent with speech may interfere with and/or suppress an individual's ability to quickly and fluently initiate and develop speech-language planning and production.

## The Present Study

This study investigated whether skin conductance was associated with developmental trajectories of stuttering persistence versus recovery. Specifically, we measured skin conductance at baseline and during a stressful picture naming task (for details pertaining to this protocol, see Zengin-Bolatkale et al., 2015). The measurements took place at three time points over a two-year period for children classified as (1) children who stuttered and recovered (recovered group), (2) children who stutter and persist (persisting group) and (3) children who do not stutter (non-stuttering group). This longitudinal approach allowed measurement of skin conductance at two points in the early development of childhood stuttering: (1) an *initial time point* close to the onset of stuttering when children classified as recovered and persisting were both stuttering, and (2) a *final time point* when only children classified as persisting were stuttering. In part, our hypotheses were designed to test whether emotional reactivity, as indexed by skin conductance level, is associated with stuttering persistence (e.g., do persisting children exhibit higher skin conductance levels compared to recovered children?) or the diagnosis of stuttering (e.g., do both persisting and recovered children exhibit higher skin conductance level than non-stuttering group?). Based on emerging findings (e.g., Ambrose et al., 2015), we hypothesized that emotional reactivity, indexed physiologically by skin conductance level, would be associated with stuttering persistence. To determine support for our speculation we empirically studied three research issues and associated testable hypotheses.

The first issue addressed whether skin conductance level (SCL) in response to stress at the *initial time point* is associated with stuttering chronicity (i.e., persistence vs. recovery). We hypothesized that persisting children, compared to recovered and non-stuttering children, would exhibit higher SCL in response to a stressful picture naming task at their initial ($1^{st}$) time point (i.e., prior to stuttering resolution for recovered children).

The second issue addressed whether SCL in response to stress at the *final time point* is associated with stuttering chronicity (persistent vs. recovered patterns). We hypothesized that persisting children, compared to recovered and non-stuttering children, would exhibit higher SCL in response to a stressful picture naming task at their final ($3^{rd}$) time point (i.e., after stuttering resolution for children who recover).

The third issue addressed whether changes in SCL in response to stress are associated with changes in stuttering frequency across the approximately 2-year period (16-32 months) for all children who stutter (i.e., persisting + recovered). We hypothesized that for the persisting and recovered groups combined, change in stuttering frequency from the initial ($1^{st}$) time point to the final ($3^{rd}$) time point would be correlated with change in SCL during the stressful picture naming task from initial time point to the final time point.

# Methods

## Participants

Data from 206 children were collected as part of a large-scale, longitudinal investigation of linguistic and emotional contributions to childhood stuttering. Following the application of the inclusion criteria described below, skin conductance data during the stressful rapid picture naming task were available for 49 children for three complete visits (each 7-10 months apart). These 49 participants were classified as belonging to one of the three following groups: (1) 9 children with persisting stuttering (1 female), (2) 23 recovered children (6 females) and (3) 17 non-stuttering children (6 females)[1]. All 49 children were monolingual Standard American English speaking and between 3 years 0 months to 6 years 11 months of age.

The Vanderbilt University's Institutional Review Board approved the present study's protocol. Informed consent by parents and assent by children were obtained. Participants

[1]Present study's participants' data were also in a previous study (Zengin-Bolatkale et al., 2015), with this previous empirical study involving different hypotheses, with none overlapping with the present study.

were recruited through advertisements in a local, free, monthly parent-oriented magazine, referrals from the Vanderbilt University speech clinic and other health professionals, and self-referrals. They voluntarily participated and received monetary compensation for their time and travel. Participants had no known or reported hearing, neurological, developmental, academic, intellectual or emotional problems, or speech and/or language disorders other than stuttering.

## Inclusion and Classification Criteria

To reduce the possibility of confounds with clinically significant speech-language and hearing concerns, participants' articulation, receptive and expressive language skills, and hearing abilities, were assessed using standardized measures. Specifically, the "Sounds in Words" subtest of the Goldman-Fristoe Test of Articulation-2 (GFTA-2; Goldman & Fristoe, 2000) measured articulation; the Peabody Picture Vocabulary Test-Fourth Edition (PPVT-4 Dunn & Dunn, 2007) assessed receptive vocabulary; the Expressive Vocabulary Test (EVT; Williams, 1997) evaluated expressive vocabulary; and the Test of Early Language Development-3 (TELD-3; Hresko, Reid, & Hammill, 1999) measured receptive and expressive language abilities. Children who scored below the 16th percentile (i.e., approximately one standard deviation below the mean) on any test were not included in the current study. In addition, bilateral pure tone hearing screenings (i.e., 25 dB HL at pure-tone frequencies of 1000 i.e., 25 dB HL at pure-tone frequencies of 2000, and 4000 Hz) were conducted. Participants were excluded if they did not perform within normal limits on this hearing screening (American Speech-Language-Hearing Association [ASHA, 1990]).

**Children who stutter—**At study entry, all 49 children participated in a diagnostic visit. Children were classified as children who stutter if the child (a) exhibited at least 3 stuttered disfluencies per 100 words in a 300-word conversational play sample and (b) scored at least 11 on the Stuttering Severity Instrument (SSI; Riley & Bakker, 2009). After the initial time point, children participated in an additional 2-3 visits over 16-32 months. Children who stutter were further divided into a persisting group and a recovered group.

**Persisting group—**Children were classified as persisting if at both the initial and final time point they (a) exhibited at least 3 stuttered disfluencies per 100 words in a 300-word conversational speech sample at each visit and (b) scored at least 11 on the SSI at each visit and (c) showed parental concern of continued stuttering at the final time point. Nine children were classified as persisting. As per parent report, 2 children in the persisting group received treatment for stuttering during the study.

**Recovered group—**Children were classified as recovered if at the final but not the initial time point they (a) exhibited below 3 stuttered disfluencies per 100 words in two 300-word conversational speech samples spaced 1-2 weeks apart approximately two years after study entry and (b) scored 10 or below on the SSI and (c) had no parental concern of continued stuttering at the final time point. Twenty-seven children were classified as recovered. As per parent report, 2 children in the recovered group received treatment for stuttering during the study.

**Non-stuttering group—**Children were classified as non-stuttering if at both the initial and final time points they (a) exhibited below 3 stuttered disfluencies per 100 words in a play sample and (b) scored 10 or below on the SSI-3. These children were also followed — after the initial time point — for at least 2 additional time points over a period of 16-32 months. Seventeen children met criteria for classification as non-stuttering at all three complete visits, with none of the 17 non-stuttering children receiving therapy for stuttering.

Lastly, in the present study, time since onset information for children who stutter was obtained from parents using a bracketing procedure described by Yairi and Ambrose (1992) and Anderson et al. (2003).

## Procedure

Participants and their parents visited our lab every 7-10 months (when available) spanning a period of 16-32 months. At each visit, one examiner engaged the child in play and collected a 300-word conversational speech sample before administering standardized speech and language tests and presenting the stressful-picture naming task. In an observation room, a second examiner conducted the parent interview.

**Stressful Picture-Naming Task—**As described in a previous empirical study (Zengin-Bolatkale et al., 2015), the essential aspects of the stressful picture-naming task are as follows. At each time point, participants completed a task that involved 30 picture cards displaying simple objects or actions selected from the Peabody Picture Vocabulary Test-Fourth Edition (PPVT-4; Dunn & Dunn, 2007). A one-minute baseline skin conductance measure was collected at the beginning and at the end of the task, during which the participant was asked to sit still and wait for the examiner "to finish some paperwork." Following the baseline skin conductance measure, participants were instructed to name each picture as rapidly as possible. The purpose of these instructions was to attempt to impose communicative, temporal, and/or interpersonal stress/pressure on participants, similar to what they might experience in communicative settings. Consistent with the purpose of the task, the examiners provided no positive feedback to the participants, but encouraged them to "go faster" sporadically during the task. The examiner placed each picture on the table, one at a time. As soon as the examiner placed a picture on the table, the next card was held up in the other hand, with the back of it facing the participant. As soon as the participant named the card on the table, the examiner slapped the next card down on the table. At the end of the task, each participant was praised and asked to sit still and wait while post-task skin conductance was measured.

**Equipment, data acquisition and processing—**Participants' bipolar electrodermal activity (i.e., SCL in this context) and acoustic recordings were acquired using a microphone and Biopac MP150 system (Biopac Systems, USA) connected to a Macintosh computer. Data were recorded using Acknowledge software (ver. 4.1 for Mac, Biopac). SCL was recorded with a pair of Ag-AgCl electrodermal electrodes placed on the palmar surface on the distal phalange of the index and the fourth fingers of the participants' left hand. There was no special preparation of participants' palmar surface because this is known to reduce the natural conductive properties of the skin (Dawson et al., 2007). Following data collection procedures recommended by Boucsein (1992), Figner and Murphy (2011), and Dawson et al. (2007), skin conductance was sampled at 500 Hz, with the gain set at 10 μS/V and a low-pass filter at 1 Hz. The testing room temperature ranged from 18 to 24 ○C. The SCL data were expressed in microSiemens (μS), which are units of measurement for conductance. Skin conductance was synchronized with each participant's acoustic recordings during picture naming. In very few occasions (for less than 5% of the total data), skin conductance data collection failed (e.g., if participants took off or pulled the electrodes during data collection), which resulted in short intervals of missing data. Therefore, during data processing, each data file was inspected to identify whether there were any intervals of missing data. In the case of intervals of missing data, we employed a linear interpolation technique (i.e., the "Connect Endpoints" math function of the Biopac Acknowledge 4.1 software) to estimate the missing data. No more than 5% of the total data was estimated using this procedure. Following correction, skin conductance data were smoothed by applying a 125 Hz filter (i.e., upper cutoff at 125 Hz) to remove artifacts due to noise or sudden deflections. Following these procedures, a mean tonic SCL value for pre-task baseline, picture naming, and post-task baseline were calculated separately, after phasic responses were removed from the signal.

## Statistical Analyses

All statistical analyses were done using Statistical Package for Social Sciences Statistics (SPSS Statistics, IBM Corp) version 24.

The first hypothesis (i.e., at the initial time point, persisting children, compared to recovered and non-stuttering children, would exhibit higher SCL in response to a stressful picture naming task) was assessed using a one-way analysis of covariance (ANCOVA) with SCL during stressful picture-naming task at initial time point as the dependent variable, and Talker Group as the independent variable. Covariates for this analysis were pre-task baseline SCL, stuttering frequency during stressful picture naming task, chronological age (in months) at initial time point and gender (rationale for all covariates used will be described at the end of this Statistical Analyses section).

The second hypothesis (i.e., at the final time point, persisting children, compared to recovered and non-stuttering children, would exhibit higher SCL in response to the task) was assessed using a one-way ANCOVA with SCL during stressful picture-naming task at final time point as the dependent variable, and Talker Group as the independent variable. Covariates for this analysis were pre-task baseline SCL, stuttering frequency during stressful picture naming task, chronological age (in months) at final time point and gender were covariates for this analysis.

The third hypothesis (i.e., for both persisting and recovered group combined, change in stuttering frequency from the initial time point to the final time point were correlated with change in SCL during the task from initial time point to the final time point.) was assessed using linear regression. First, to index "reactivity to stressful picture naming task", we calculated residualized change scores for each participant's SCL during the task by using baseline SCL, stuttering frequency during stressful picture naming task, age, and gender as covariates. Next, we computed variables to index (1) amount of *change* in SCL during the stressful picture-naming task (i.e., using residualized change scores for SCL at each time point) from the initial time point (T1) to the final time point (T3) and (2) the amount of *change* in stuttering frequency from the initial time point (T1) to the final time point (T3). Last, we used linear regression to assess the relation between SCL change score (T3-T1) and stuttering frequency change score (T3-T1).

### Rationale for covariates

**Pre-task baseline SCL—**According to Wilder's law of initial values, pre-task baseline SCL values could influence subsequent SCL scores (Wilder, 1962). Further, it is recommended that baseline SCL values be taken into consideration because there is inter-individual variance in SCL due mainly to physiological variables unrelated to psychological processes (e.g., thickness of the skin in recording area) (Dawson et al., 2007; Lykken & Venables, 1971). Therefore, to quantify change in emotional *reactivity* from baseline, as indexed by SCL, and to minimize inter-individual variance and relation between baseline SCL and subsequent SCL, pre-task baseline SCL was used as a covariate for all analyses.

**Chronological age—**Previous studies reported that chronological age is important for analyses of SCL (e.g., El-Sheikh, 2005, 2007; El-Sheikh & Arsiwalla, 2011; Venables & Mitchell, 1996). More recently, a cross-sectional study of children who do and do not stutter during a stressful picture-naming task reported significant between-group differences in SCL as a result of chronological age (Zengin-Bolatkale et al., 2015).

**Gender—**Gender differences have been reported in autonomic arousal and emotional responses of children and adults (e.g., Arnold et al., 2014; Boucsein, 1992; Chentsova-Dutton & Tsai, 2007).

**Stuttering Frequency during Picture-Naming Task—**Lastly, to account for the possible impact of stuttering events on SCL, stuttering frequency during stressful picture naming task was a covariate in all analyses.

## Results

### Descriptive Statistics

Table 1 shows participants' demographic, speech fluency, speech and language, attitudes towards talking and temperament data at the initial time point. One-way ANOVA assessed between group differences in age, speech fluency, and speech and language scores. Chi-square analysis assessed between-group differences in gender.

**Demographic Variables—**There was a significant difference in chronological age among the three groups. Follow-up analyses comparing recovered and persisting groups indicated no between-group differences in chronological age ($F(1, 30)= .578$, $p=.453$). The mean chronological age for non-stuttering group was significantly higher than the persisting group ($F(1, 24)= 5.219$, $p=.031$) and the recovered group ($F(1, 38)=5.373$, $p=.026$). Therefore, and as stated above, age was included as a covariate in the statistical analyses for the main hypotheses. Despite non-significant between-group differences in gender, it was also included as a covariate in analyses of the main hypotheses.

**Speech Fluency During Conversation—**At the initial time point, as would be expected from inclusion criteria, the persisting group and the recovered group exhibited higher frequency of stuttered disfluencies and higher SSI scores than the non-stuttering group. Follow-up analyses comparing the recovered and persisting groups indicated no between-group differences in stuttering frequency ($F$(1, 30)= .025, $p$=.876) or SSI-3 scores ($F$(1, 30)= .337, $p$=.566). Last, there was no between-group difference in time since onset for the recovered group and the persisting group.

**Speech Fluency During Stressful Picture-Naming Task—**At the initial time point, there were no significant between-group differences among persisting ($M$= 2.07, $SD$= 2.08), recovered ($M$=2.38, $SD$=3.89), and non-stuttering group ($M$= 1.61, $SD$= 1.63) in stuttering frequency during stressful picture naming task $F$(2, 45)= .335, $p$=.717. Further, at the final time point, there were no significant between-group differences among persisting ($M$= .53, $SD$= 1.10), recovered ($M$=.72, $SD$=1.29), and non-stuttering group ($M$= .99, $SD$= 1.42) in stuttering frequency during stressful picture naming task $F$(2, 46)= .42, $p$=.66.

**Speech and Language—**No between-group differences were found in articulation, receptive and expressive vocabulary, and receptive and expressive language for the non-stuttering, recovered and persisting groups.

**Temperament and Attitudes Towards Talking—**There were no between-group differences in temperament, as indexed by parent reports to the Children's Behavioral Questionnaire (CBQ, Rothbart, Ahadi, & Hershey, 1994; Rothbart, Ahadi, Hershey, & Fisher, 2001). Likewise, there was no between-group difference in attitudes towards talking, as indexed by children's reports to a Communication Attitude Test for Preschool and Kindergarten Children Who Stutter (KiddyCAT, Vanryckeghem & Brutten, 2007).

### À Priori Hypotheses

(Hypothesis 1): Persisting children compared to recovered and non-stuttering children would exhibit higher SCL in response to a stressful picture naming at the initial (1$^{st}$) time point

One-way ANCOVA assessed between group differences in SCL during the stressful picture-naming task at the initial time point. Figure 1 shows estimated marginal means for SCL at the initial time point for persisting, recovered, and non-stuttering groups. There was a significant main effect for Talker Group ($F$(2, 42)= 3.657, $p$= 0.035,, $\eta_p^2$=.151). Follow-up simple comparisons indicated that the persisting group ($M$= 14.75, $SE$=.68) had significantly higher SCL than the recovered group ($M$=12.73, $SE$=.43) ($F$(1, 25)= 6.178, $p$=.020, $\eta_p^2$=.198), but was only marginally different from the non-stuttering group ($M$= 13.29, $SE$=.59) ($F$(1, 20)= 4.091, $p$=.057, $\eta_p^2$=.170). There was no difference in SCL during the picture naming task between recovered and non-stuttering groups ($F$(1, 33)= .192, $p$=.664, $\eta_p^2$=.006). Therefore, the first hypothesis was partially supported.

Hypothesis 2: Persisting children compared to recovered and non-stuttering children would exhibit higher SCL in response to a stressful picture naming at the final (3rd) time point

One-way ANCOVA assessed between-group differences in SCL during the task at the final time point. The main effect for Talker Group was not significant ($F(2, 42)= .316$, $p= 0.731$, $\eta_p^2=.015$). Thus, there were no significant differences in SCL among persisting ($M=15.73$, $SE=.92$), recovered ($M=15.66$, $SE=.57$), and non-stuttering ($M=14.98$, $SE=.69$) groups at the final time point. Therefore, Hypothesis 2 was not supported.

Hypothesis 3: For both persisting and recovered groups combined, change in stuttering frequency from the initial (1st) time point to the final (3rd) time point would be correlated with change in SCL during the task from initial time point to the final time point.

This analysis was performed by first calculating change scores for SCL and stuttering frequency from first time point to final time point (T3-T1) and then by investigating the relation between the two variables using linear regression. For both persisting and recovered groups combined, the correlation between change in stuttering frequency (T3-T1) and change in SCL (T3-T1) was not statistically significant $r= .138$, $p=.496$. Therefore, Hypothesis 3 was not supported.

### Ancillary Analyses

Table 2 shows speech fluency and SCL measures for each of the 3 talker groups across the three time points (T1, T2, T3). To determine how each of the three groups' SCL responses changed at each of the three time periods, a mixed-model ANOVA was conducted with Talker Group as the independent variable (3: persisting, recovered, non-stuttering) and SCL during stressful picture naming task as the dependent variable, and Time Points as a repeated measure (3: T1, T2, T3). Covariates were gender, age, stuttering frequency during picture naming task, and baseline SCL at each time point. Findings indicated that there were no significant main effects for Talker Group $F(2, 127.04)= .541$, $p= .583$ and Time Point $F(2, 98.452)= .117$, $p= .89$. Further, the interaction effect for Talker Group × Time Point was not significant $F(4, 81.379)= 1.813$, $p= .134$. These findings suggested that the groups did not significantly differ in SCL change between time points.

## Discussion

### Summary of Main Findings

The present study resulted in one main finding. This finding suggests that the persisting, when compared to recovered group exhibited significantly higher sympathetic arousal (i.e., higher SCL) during the stressful-picture naming task at the initial time point. Below we will discuss the implications of this finding.

## Implications of Main Findings

**Heightened reactivity to stress: A possible marker for persistence in childhood stuttering—**Sympathetic arousal, as indexed by SCL, has been reliably used as a marker for emotional reactivity (e.g., Boucsein, 1992; Dawson et al., 2007; Porges, 2007; Sequeira et al., 2009). Further, increases in SCL during challenge or stressful conditions have been associated with heightened emotional reactivity and stress (e.g., Fabes et al., 1994; Fowles et al., 2000). Thus, present findings indicate that children for whom stuttering persists, relative to those who recover, exhibited greater emotional reactivity during the stressful picture-naming task at initial testing. At that testing both groups stuttered at a comparable rate (i.e., several months before their classification as persisting or recovered was determined). Therefore, one might speculate that there might be increased risk for persistence for CWS who exhibit heightened emotional reactivity to stressful events early in the development of their stuttering.

In support of this speculation, emotions have been linked to stuttering persistence in a recent longitudinal report (Ambrose et al., 2015). Specifically, Ambrose and colleagues (2015) reported that based on caregiver reports of temperament at initial testing or time point, the persistent group exhibited greater negative affectivity than recovered group and non-stuttering groups. Additionally, Ambrose et al.'s analyses of the sub-scales associated with the negative affectivity factor indicated that children in the persistent group were temperamentally more fearful and less soothable than children in the recovered or non-stuttering groups. Although the relation between emotions and stuttering chronicity has yet to be comprehensively studied, the findings of the Ambrose et al. study are consistent with the present study's main finding that the persisting group, compared to the recovered group, showed higher SCL (i.e., higher emotional reactivity) during the stressful picture naming task at the initial time point. Such findings can be explained by at least three potential accounts.

First, the persisting group may exhibit greater temperamental tendencies to be reactive during stressful situations. This account is consistent with Ambrose et al.'s (2015) findings that aspects of persisting children's temperament may contribute to them reacting differently (and more negatively) to unpleasant or stressful situations than recovered or non-stuttering children.

Second, and alternatively, it could be speculated that children in the persisting group showed heightened sympathetic arousal during the stressful picture-naming task due to their experiences with stuttering. For several reasons, however, experience with stuttering does not appear to be a robust account for our finding that persisting children exhibited greater sympathetic arousal than children who recovered. First, if this account were the case, then one would expect that children in the recovered group would also show heightened sympathetic arousal at the initial time point, which we did not find. Second, at the initial time point, there were no significant differences between the two groups in chronological age or parental reports of time since onset of stuttering. Third, both persisting and recovered groups were stuttering at comparable rates at the initial time point. Even so, one cannot categorically rule out the influence of experience with stuttering on children's emotion.

However, neither can one rule out the possibility that children in the persisting group, relative to those in the recovered group, have a greater proclivity to be highly sensitive to their experiences with stuttering from onset of stuttering onward and thus develop greater reactivity to stressful speaking situations.

Finally, one might conjecture that there is a ‘sensitive period’ early in the course of the development of stuttering, during which concomitant heightened emotional reactivity might increase the risk of persistence. According to a popular theory of spoken language development (Locke, 1992; 1994), linguistic capacity develops gradually and sequentially at critical time periods. The onset of stuttering coincides with a sensitive period between the ages of 2 and 4 during which children make significant gains in receptive and expressive speech and language skills (e.g., Ratner, 1997, Yairi, 1983). Similarly, between ages 2 and 5, children typically develop a broader range of abilities to regulate their own emotions (e.g., Kopp, 1989; Thompson, 1994). One might speculate that heightened emotional reactivity during this sensitive period might cause speech and language system to develop in a different way relative to typically developing peers. According to this speculation, if a child who stutters also has higher emotional reactivity relative to peers between the ages of 2 and 4 years, he or she might have a greater risk of persisting in stuttering.

Consistent with such speculation is the fact that at the present study’s initial time point, children in both persisting and recovered groups were between the ages of 3 and 4 and that children who persisted, relative to those who recovered, exhibited greater SCL during stressful picture naming task at initial time point, but not the final time point. Similarly, Ambrose et al. (2015) reported that differences between persistent and recovered group’s temperamental development were most apparent at the initial time point. Therefore, higher sympathetic arousal (i.e., indexing higher emotional reactivity) during stressful speaking situations measured as close to the onset of stuttering as possible (i.e., as close to the “sensitive period” as possible) might be one useful early marker for persistence in stuttering. At present, support for or refutation of the three accounts discussed above must await the findings of future studies. Such empirical studies should help determine the potential long-term impact of high emotional reactivity during sensitive period of stuttering and speech development, as well as the relative import of the “nature” (temperamental) and “nurture” (stuttering experience) contributions to emotional processes in children who stutter.

## Caveats

One limitation of the present study was the relatively small sample size of participants, particularly for children whose stuttering persisted, something to be expected based on typical persistence and recovery rates (for review, see Yairi & Ambrose, 2013). Whatever the case, the relatively smalls sample sizes, particularly for children who persist, may be a limiting factor for broadcast generalizations of findings for persistent, recovered and non-stuttering children (although cautious generalization would seem plausible).

A second concern, also related to sample size, is the possible use of scatterplots to determine whether outliers may have impacted reported central tendencies. Such usage is challenging for at least two reasons: (1) standard errors of the means are quite comparable at each time point across groups, and, perhaps most importantly, (2) the central tendencies reported had a number of covariates associated with them (e.g., baseline SCL, age, gender). Scatterplots would neither show nor reflect the influence of such covariates, rendering the use of scatterplots of dubious value for interpreting reported central tendencies (associated with H1-3).

A third concern with the present findings is that the SCL of the persisting group was marginally different than that of the non-stuttering group at p=0.057 level. This finding undoubtedly was impacted by both sample size and hence issues with statistical power. Further discussion regarding the absolute lack of statistical significance would be, in these authors' opinion, less than meaningful. Future empirical studies with increased statistical power are necessary to determine whether or not there are significant differences in SCL between persisting and non-stuttering groups.

A fourth limitation is that the present study assessed emotional reactivity during a stressful picture-naming task, eliciting one-word response, rather than using a conversational speaking task or a narrative. Clearly, conversational or narrative, when compared to one-word, tasks involve linguistically more complex productions such as phrases and sentences, and would seem ecologically closer to the speaking conditions during which stuttering is most apt to occur. Indeed, young children have been reported to be fairly fluent during speaking conditions that involve single-word responses such as "yes" "no" or as in a picture-naming task when compared to running speech in a conversational sample (Yaruss, 1998; Lee, 1974). Consistent with this observation, present results indicate minimal stuttering frequency during the picture-naming task. Such infrequent stuttering during the picture-naming task appreciably reduced our ability to determine the association between stuttering frequency and SCL during the task.

Another limitation of the present study is that participants completed the same stressful picture-naming task at every time point. It seems hard to give such a concern much credence given the fact that there were approximately 8 months separations between each participant's experience with the task. Nevertheless, it is possible participants got accustomed to the task in their subsequent visits and did not perceive the task to be as challenging as they did at the initial time point (of course, this would be a "constant error" because it was the same for all three talker groups). Therefore, it might be prudent for future studies using the present paradigm to make age-appropriate adjustments such as changing the delivery of degrees of stress/time pressure, or the length of the task.

Lastly, it is tempting to compare the present SCL findings to those previously reported in 2015 by these authors. Doing so, however, would involve comparison of past *cross-sectional* (Zengin-Bolatkale et al., 2015) to present *longitudinal* findings. In the earlier, cross-sectional study there were significant differences in SCL between 3-year-olds CWS and CWNS. In the present longitudinal study, however, there were significant differences in SCL between children who persisted versus recovered, the latter longitudinally sampled information not available in the earlier cross-sectional study (although the past cross-sectional finding of the influence of age on SCL lead to use of chronological age as a covariate in all of the present study's inferential analyses). Thus, it is likely that the relative contributions of chronological age and persistence/recovery from stuttering to SCL are somewhat entangled, with any attempts at disentanglement requiring further empirical study.

## Conclusion

The present study's findings provide support for the notion that sympathetic arousal during a stressful speaking task measured close to the onset of stuttering might be a possible marker for persistence or recovery. To the current authors' knowledge, the present empirical study is one of the first of its kind to longitudinally investigate physiological associates of emotion and its role in persistence and recovery from childhood stuttering. Therefore, future replications are essential. Accumulating knowledge from this and subsequent studies will aid in developing a better understanding of the association between emotion and childhood stuttering and its role in persistence and recovery. Whether the directionality of effect between emotion and childhood stuttering is eventually shown to be (1) emotion → stuttering, (2) emotion ← stuttering or even (3) emotion ⇔ stuttering, present findings and those of others (e.g., Eggers et al., 2010; Jones et al., 2014; Ntourou et al., 2013) indicate that emotion should be considered in any comprehensive account of childhood stuttering.

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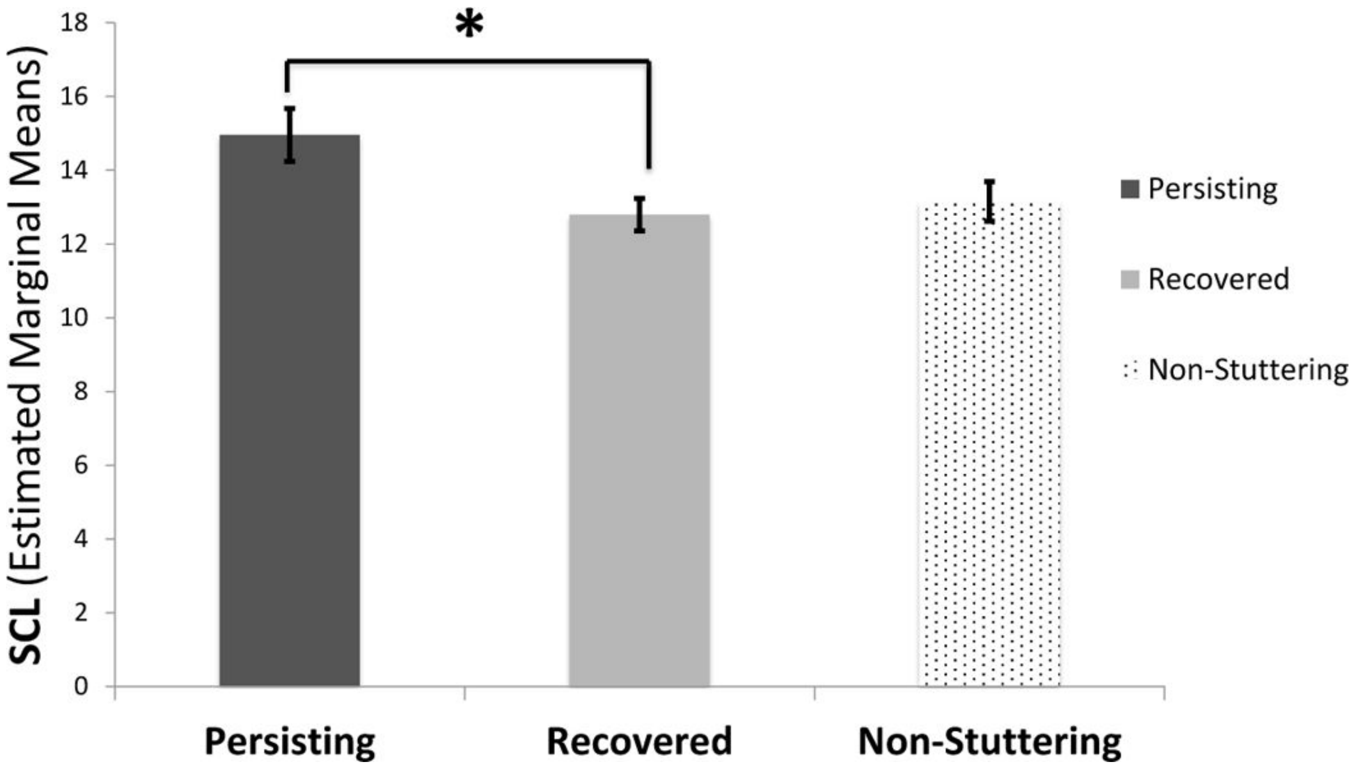


**Figure 1.**

SCL (estimated marginal means) during stressful-picture naming task at initial time point for children in persisting, recovered and non-stuttering groups.

*Note*: Estimated marginal means represent SCL during the stressful picture naming task with age, gender, and baseline SCL as covariates. Error bars represent standard errors. * indicates p< .05

**Table 1**

Demographic, speech fluency, speech-language, attitudes toward talking and temperament characteristics of the persisting, recovered and non-stuttering group at the initial visit. (T1)

| Variable | Persisting (n=9) | Recovered (n=23) | Non-Stuttering (n=17) | F (df) | Wald $\chi 2$ (df) | p | Effect Size |
|---|---|---|---|---|---|---|---|
| Chronological age (mos.) | 45.11 (6.23) | 47.109 (6.84) | 53.06 (9.27) | 4.142 (2,46) | | 0.022 | 0.153 |
| Gender | 1 female | 6 females | 6 females | | 1.77 (2) | 0.413 | 0.235 |
| **Speech Fluency Measures** | | | | | | | |
| Stuttering Frequency (%) | 6.51 (4.52) | 6.25 (4.37) | 1.31 (.82) | 10.79 (2,46) | | 0.0001 | 0.319 |
| SSI-3 Total Score | 16.56 (7.65) | 15.3 (4.43) | 6.71 (2.08) | 21.32 (2,46) | | 0.0001 | 0.481 |
| Time since onset (mos.) | 11.38 (7.62) | 9.46 (5.66) | – | .399 (1, 17) | | 0.536 | 0.023 |
| **Speech and Language Testing** | | | | | | | |
| GFTA-2 | 109.44 (8.05) | 111.87 (7.38) | 108.76 (8.55) | .823 (2,46) | | 0.445 | 0.035 |
| PPVT-4 | 120.67 (16.12) | 116.35 (12.1) | 120.53 (12.55) | .643 (2,46) | | 0.531 | 0.027 |
| EVT-2 | 121.56 (12.11) | 118 (11.25) | 120.35 (10.7) | .392 (2,46) | | 0.678 | 0.017 |
| TELD-3 Receptive | 116.67 (14.46) | 122.86 (15.35) | 115.59 (14.4) | 1.27 (2,44) | | 0.292 | 0.054 |
| TEL-3 Expressive | 109.67 (12.47) | 116.67 (16.79) | 106.29 (11.98) | 2.51 (2,44) | | 0.092 | 0.103 |
| **Attitudes toward Talking (KiddyCAT) and Temperament (CBQ)** | | | | | | | |
| KiddyCAT | 3.89 (2.76) | 2.95 (2.15) | 2.94 (2.25) | .609 (2,46) | | 0.548 | 0.026 |
| CBQ Negative Affectivity | 3.82 (.37) | 3.96 (.43) | 3.69 (.51) | 1.675 (2,44) | | 0.199 | 0.071 |
| CBQ Surgency | 4.75 (.704) | 4.83 (.86) | 4.87 (.704) | 0.067 (2,44) | | 0.935 | 0.003 |
| CBQ Effortful Control | 4.50 (.76) | 4.66 (.46) | 4.93 (.61) | 1.98 (2,44) | | 0.151 | 0.082 |

*Note*. SSI-3 – Stuttering Severity Instrument – Third Edition; GFTA-2= Goldman Fristoe Test of Articulation-Second Edition; PPVT-4 = Peabody Picture Vocabulary Test- Fourth Edition; EVT-2 = Expressive Vocabulary Test – Second Edition; TELD-3= Test of Early Language Development–Third Edition; KiddyCAT= Communication Attitude Test for Preschool and Kindergarten Children Who Stutter; CBQ= Children's Behavior Questionnare. Effect size for gender is measured using Cohen's d; all other effect sizes are measured using partial eta squared (ηp2). There were missing data for two recovered participants for the following variables: TELD-3 Receptive and Expressive Time since onset data at initial visit was not availabe for 2 children in persisting group and 11 children in recovered group. CBQ data were not available for 1 child in the recovered group and 1 child in the non-stuttering group.

## Table 2

Speech fluency and Skin Conductance Level measures for the persisting, recovered, and non-stuttering groups for each of the 3 time points (T1, T2, T3).

| | Persisting (n=9) | | | Recovered (n=23) | | | Non-Stuttering (n=17) | | |
|---|---|---|---|---|---|---|---|---|---|
| **Variable** | **T1** | **T2** | **T3** | **T1** | **T2** | **T3** | **T1** | **T2** | **T3** |
| **Speech Fluency Measures (during conversation)** | | | | | | | | | |
| Stuttering Frequency (%) | 6.51 (4.52) | 3.67 (1.1) | 3.4 (.77) | 6.25 (4.37) | 2.84 (1.9) | 1.03 (.77) | 1.31 (.82) | 1.12 (.7) | .88 (.75) |
| SSI-3 Total Score | 16.56 (7.65) | 12.22 (2.54) | 12.33 (2.91) | 15.3 (4.43) | 9.91 (3.9) | 6.3 (1.85) | 6.71 (2.08) | 5.8 (1.65) | 5.65 (1.9) |
| **Skin Conductance Level (during Stressful Picture-naming Task)** | | | | | | | | | |
| Skin Conductance Level | 15.09 (.71) | 12.36 (.98) | 15.73 (.92) | 12.88 (.44) | 13.17 (.59) | 15.66 (.57) | 13.19 (.53) | 12.96 (.64) | 14.98 (.69) |

Note. For speech fluency measures, the values indicate mean (SD); for skin conductance level measures, the values indicate estimated marginal means (SE).