HHS Public Access

Author manuscript

J Commun Disord. Author manuscript; available in PMC 2024 March 01.

Published in final edited form as:

J Commun Disord. ; 102: 106304. doi:10.1016/j.jcomdis.2023.106304.

A Study of Respiratory Sinus Arrhythmia and Stuttering Persistence

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Abstract

Introduction: The present study investigated potential differences in respiratory sinus arrhythmia between preschool-age children with persisting stuttering, children who recovered from stuttering, and children who do not stutter.

Methods: Participants were 10 children with persisting stuttering (persisting group), 20 children who recovered from stuttering (recovered group), and 36 children who do not stutter (non-stuttering group). Participants viewed a neutral video clip to establish a pre-arousal baseline and then viewed two emotionally-arousing video clips (positive and negative, counterbalanced). Age-appropriate speaking tasks followed each of the video clips (post-baseline, post-positive, and post-negative). Respiratory sinus arrhythmia (RSA), an index of parasympathetic nervous system activity, was measured during the video clips and subsequent speaking tasks.

Results: First, the persisting group, recovered group, and non-stuttering group did not significantly differ in baseline RSA. Second, during the emotionally-arousing video clips, there was a significant *group × condition interaction*, with the recovered group exhibiting significantly lower RSA in the positive than negative condition, and the non-stuttering group exhibiting

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Author Statement

Dillon G. Pruett: Conceptualization, Formal Analysis, Investigation, Data Curation, Writing - Original Draft, Writing - Review & Editing **Stephen W. Porges:** Software, Methodology, Writing - Review & Editing **Tedra A. Walden:** Conceptualization, Methodology, Resources, Writing - Review & Editing **Robin M. Jones:** Conceptualization, Methodology, Formal Analysis, Validation, Investigation, Resources, Writing - Review & Editing, Supervision, Project Administration

Vanderbilt University’s Institutional Review Board approved the protocol presented in this study. Informed consent by parents and assent by children were obtained.

significantly higher RSA in the positive than negative condition. Third, in the narrative tasks, there was a significant *group × condition interaction*, with a greater difference in RSA between the post-baseline speaking task and the post-positive and post-negative speaking tasks for the persisting compared to the non-stuttering group. Lastly, a follow-up analysis indicated that the recovered and nonstuttering groups, compared to the persisting group, exhibited significantly greater RSA during the baseline (neutral) condition compared to the post-neutral narrative task.

Conclusions: Findings provide a physiological perspective of emotion within children who stutter and persist and children who stutter and recover. Future investigations with larger sample sizes and diverse methodologies are necessary to provide novel insights on the specific emotion-related processes that are potentially involved with persistence of stuttering in young children.

Keywords

developmental stuttering; respiratory sinus arrhythmia; emotion regulation

Introduction

Developmental stuttering is a neurodevelopmental disorder that begins in early childhood and is characterized by repetitions and prolongations of speech sounds and syllables (Yairi & Ambrose, 2013). About 80–85% of children who stutter spontaneously recover without formal treatment; however, the remaining 15–20% stutter into adulthood (Yairi & Ambrose, 1999). For those who continue to stutter, differences in speech can dramatically impact emotional and social wellbeing on a daily basis (Beilby et al., 2012). Despite a growing empirical knowledge base of contributors to developmental stuttering, there is a need for a multifactorial understanding of stuttering persistence consistent with theoretical speculation on the precipitating and exacerbating factors of stuttering (Conture & Walden, 2012; Smith & Weber, 2017). Identifying the factors associated with stuttering recovery may inform our understanding of the pathogenesis of stuttering and significantly help caregivers and speech-language pathologists focus resources on children at the highest risk for persistence.

Temperament, Emotion, and Stuttering

Temperament and emotion have both been implicated theoretically in the early onset and persistence of childhood stuttering (Conture et al., 2013). A number of cross-sectional investigations assessed differences in temperament and emotion between children who stutter and children who do not stutter (e.g., Anderson et al., 2003; Karrass et al., 2006; Schwenk et al., 2007), and reviews indicate that there may be an association between temperament, emotion and stuttering (Conture et al., 2013; Jones, Choi, et al., 2014; Kefalianos et al., 2012). For example, children who stutter, compared to children who do not stutter, exhibit: (1) less adaptability (e.g., Anderson et al., 2003; Schwenk et al., 2007); (2) more negative affect/mood (e.g., Jones, Choi, et al., 2014; Ntourou et al., 2013); (3) higher emotional reactivity (e.g., Jones et al., 2014; Karrass et al., 2006; Zengin-Bolatkale et al., 2015); and (4) lower emotion and attention regulation (Eggers et al., 2013; Jones, Buhr, et al., 2014; Karrass et al., 2006).

Fewer longitudinal studies assessing emotion and temperament in children who stutter have been conducted. One relatively recent longitudinal study followed children and assessed epidemiology, speech motor skills, language, and temperament over the course of four years. Results indicated that children with persistent stuttering, compared to children who stuttered and recovered and children who do not stutter, were rated by caregivers as significantly higher in negative affectivity using the Children's Behavior Questionnaire (Ambrose et al., 2015). Related to these findings, Singer et al. (2019) demonstrated that higher negative affectivity was associated with lower receptive vocabulary for groups of children with persistent and recovered stuttering compared to children who do not stutter in a different longitudinal cohort. In an effort to understand emotion-speech interactions, Erdemir et al. (2018) investigated the impact of emotion on articulation rate, a measure of speech processes. Findings indicated that the persisting group exhibited significantly slower articulation rate while producing a narrative following exposure to negative emotion-inducing videos, which may indicate a breakdown in speech-related processes during negative emotion for these children. Furthermore, a study by Zengin-Bolatkale et al. (2018) examined sympathetic activity during a stressful speaking task. Findings indicated higher sympathetic arousal at the first time point in a longitudinal study by children with persisting stuttering versus children who eventually exhibited recovered stuttering. Another longitudinal study found that stuttering status and stuttering persistence were associated with atypical connectivity within the default mode network, the large-scale brain network of regions with highly correlated activity at rest, and its connectivity with attention, somatomotor, and frontoparietal networks (Chang et al., 2018). Results suggest that anomalous connectivity within networks supporting motor, emotion, attention, and perception may place individuals at risk for both stuttering and stuttering persistence.

Overall, cross-sectional and longitudinal studies suggest that there are salient differences in emotion and temperament in children who stutter, reinforcing the need for further exploration. To date, longitudinal work in this area has largely focused on emotional reactivity (Erdemir et al., 2018; Zengin-Bolatkale et al., 2018) and/or has employed caregiver report and behavioral measures in the study of regulation (Erdemir et al., 2018; Singer et al., 2019). Thus, the main purpose of the present study was to assess *autonomic regulation* near stuttering onset and its role in the persistence of stuttering, indexed by respiratory sinus arrhythmia (RSA). Compared to caregiver report and behavioral measures, physiological measures like RSA have only recently been employed in the study of developmental stuttering and can supplement measures that are based on more subjective judgments of emotion.

Respiratory sinus arrhythmia - Physiological measure of emotion regulation

Respiratory sinus arrhythmia (RSA) is a physiological measure indicating parasympathetic influence on the heart, mediated via myelinated vagal efferent pathways. RSA is operationally defined as heart rate variation at the approximate frequency of spontaneous respiration; while vagal influences on the heart cannot be directly measured, the amplitude of periodic changes in heart rate associated with the frequencies of spontaneous respiration index vagal influences on the heart (see Lewis et al., 2012) and serve as a proxy for systemic parasympathetic function (Berntson et al., 1993, 2007; Denver et al., 2007). Specifically, these patterns of heart rate variation can be measured via electrocardiogram as subtle changes in the timing between heartbeats, known as inter-beat-interval (IBI) variations, modulated by breathing, with longer IBIs occurring during expiration than inspiration. Respiratory sinus arrhythmia has been used in both biological and psychological studies as an index of the autonomic state underlying emotion regulation and can provide information about autonomic regulation that would be difficult to assess exclusively via observational techniques (Butler et al., 2006; Demaree et al., 2004; Heilman et al., 2008; Utendale et al., 2014).

Baseline RSA reflects parasympathetic nervous system activity at relative rest. In general, high baseline RSA is considered adaptive, with higher baseline RSA positively correlated with positive psychological traits including empathy and capacity to sustain attention (Beauchaine & Thayer, 2015). Conversely, low baseline RSA is considered less adaptive and is correlated with externalizing behavior problems and other disorders including anxiety, phobias, attention problems, and depression (for review, see Beauchaine & Thayer, 2015). Consequently, baseline RSA is often used as a measure of stress vulnerability: high baseline RSA reflects high *capacity* for emotion regulation and low baseline RSA reflects low *capacity* for emotion regulation.

In contrast to baseline RSA, RSA measured during challenges can be used to evaluate RSA regulation. Transient changes in RSA are a component of regulatory processes and are associated with cognitive, behavioral, emotional, attentional, and social challenges (Bar-Haim et al., 2004; Gentzler et al., 2009; Gilissen et al., 2007; Heilman et al., 2008; Marcovitch et al., 2010; Weber et al., 1994). In these conditions, heart rate increases and RSA decreases. Generally, greater decreases in RSA from baseline are associated with more adaptive responses during social and emotional challenges (Bar-Haim et al., 2004; Gentzler et al., 2009). However, there are cases where both low and moderate decreases in RSA have been associated with increased cognitive performance and executive functioning (Marcovitch et al., 2010; Utendale et al., 2014). Therefore, the magnitude of decrease in RSA that would promote adaptive performance may be task-dependent.

Respiratory sinus arrhythmia and stuttering.—Empirical results have shown promise for the use of respiratory sinus arrhythmia in the investigation of developmental stuttering. Results indicate that 1) children who stutter have lower baseline RSA than children who do not stutter, indicating a lower capacity for emotion regulation in response to challenge, and 2) children who stutter exhibit higher RSA during speech tasks than children who do not stutter, perhaps reflecting decreased and/or less efficient engagement during social-communication situations (Jones, Buhr, et al., 2014). Furthermore, decreased RSA from baseline has been associated with increased stuttered disfluencies for both children who stutter and children who do not stutter (Jones et al., 2017).

Whether measured by caregiver reports and behavioral observation or by physiological measures such as RSA there have been numerous empirical findings that indicate salient differences in temperament and emotion reactivity and regulation between children who do and do not stutter. Investigating RSA coupled with changes in stuttering over time may answer a crucial question: Is RSA associated with stuttering persistence or recovery?

The study addressed this question by examining RSA near stuttering onset and following children who stutter for two years. Respiratory sinus arrhythmia was measured during the study's six conditions: (1) neutral video clip (initial baseline), (2) emotionally arousing video clips (positive & negative), and (3) speaking tasks immediately following each video clip (neutral clip, emotionally arousing clips).

Based on theoretical perspectives (Walden et al., 2012) and emerging empirical evidence (Ambrose et al., 2015; Erdemir et al., 2018; Singer et al., 2019; Zengin-Bolatkale et al., 2018) that stuttering persistence may be associated with emotion and temperament, we speculated that RSA may serve as a risk factor for persistence. We hypothesized that, near stuttering onset, only the persisting group would exhibit decreased emotion regulation compared to the recovered group and the non-stuttering group. From this perspective, the previously observed cross-sectional differences in emotion regulation would be primarily driven by the persistent group, with the recovered group exhibiting emotion regulation more similar to the non-stuttering group.

Hypotheses for the present study were as follows:

Baseline

1. During the neutral baseline video viewing condition, the persisting group will exhibit a lower capacity for emotion regulation, indexed by significantly lower baseline RSA compared to the recovered and non-stuttering groups.

Emotionally Arousing Video Clips

2. While viewing the positive and negative video clips, the persisting group will exhibit lower emotion regulation, indexed by a higher level of RSA (adjusted for baseline), compared to the recovered and non-stuttering groups.

Speaking Tasks Following Emotionally Arousing Video Clips

3. During the speaking tasks following each of the video clips, the persisting group will exhibit lower emotion regulation, indexed by a higher level of RSA (adjusted for baseline), compared to the recovered and non-stuttering groups.

Methods

Participants

Two-hundred-and-six children participated in a longitudinal investigation of multifactorial contributions to early childhood stuttering. After applying the exclusion criteria described below, 66 monolingual, Standard American English-speaking children, aged 3;0 to 4;11, were included as participants in the present study[1]. The study protocol was approved by

[1]24 participants were included in both Jones et al. (2014) and the current study, representing 36% of the current study's sample. The current study categorized participants into three groups (*persisting*, r*ecovered*, and *non-stuttering*) based on longitudinal stuttering trajectory whereas Jones et al. (2014) categorized participants into two groups (*children who stutter* and *children who do not stutter*) based on initial time point findings. Therefore, the current study's hypotheses are investigating different groupings.

the university's Institutional Review Board and informed consent and assent by parents and children, respectively, were obtained.

Participants were recruited through advertisements in a free local monthly parent-oriented magazine, through referrals from a university-affiliated medical center offering pediatric speech-language pathology services, and through referrals from other health professionals. They had no known speech or language disorders other than stuttering, with typical hearing and typical neurological, intellectual, and social development. All participants were naïve to the purpose and design of the study and received compensation for time and travel.

Classification and Inclusion Criteria

Children who stutter.—The initial study visit included a diagnostic evaluation. To be classified as stuttering, participants (a) exhibited at least 3 stuttering-like disfluencies per 100 words in a 300-word conversational speech sample during play with a speech-language pathologist and (b) scored an 11 or higher on the Stuttering Severity Instrument (SSI; Riley, 1994; 2009)[2].Following the initial visit, children returned for 2–3 visits over 22–32 months. These children who stutter were further classified as *persisting* or *recovered*.

Persisting group.: Participants were classified as persisting if they (a) exhibited 3 or more stuttering-like disfluencies per 100 words in the 300-word conversational speech sample at each visit and (b) scored 11 or higher on the SSI at each visit. Ten participants were classified as persisting. During the study, 2 children in the persisting group received treatment for stuttering, per parent report. Specific treatment type was not reported. Parents were asked to describe their child's race from six categories: American Indian or Alaskan Native, Asian, Black or African-American, Native Hawaiian or Other Pacific Islander, White, or Multi-racial. Racial breakdown for the persisting group was 30% Black or African-American, 10% Multi-racial, and 60% White.

Recovered group.: Participants were classified as recovered if they (a) exhibited less than 3 stuttering-like disfluencies per 100 words in two consecutive 300-word conversational speech samples 1–2 weeks apart two years after beginning the study and (b) scored a 10 or below on the SSI. Twenty participants were classified as recovered. During the study, 6 participants in the recovered group received treatment for stuttering, per parent report. Specific treatment type was not reported. Racial breakdown for the recovered group was 5% Black or African-American, 85% White, and 10% Multi-racial.

Non-stuttering group.—Participants were classified as non-stuttering if they (a) exhibited below 3 stuttering-like disfluencies per 100 words in the initial speech sample and (b) scored a 10 or below on the SSI-3. These children were also followed for 2–3 visits over 22–32 months. Thirty-six participants were classified as non-stuttering at all visits.

[2]The Stuttering Severity Instrument is a diagnostic tool for stuttering based on syllables stuttered during speech samples, duration of stuttering moments, and concomitant behaviors. Children are given ratings and there is no non-stuttering category. The lowest severity category, very mild, corresponds to ratings of 8 or below. To minimize overlap between children who stutter and children who do not stutter in this study, children were categorized as "unclassifiable" if their stuttering frequency and SSI score were incongruent. The 3rd and 4th editions of the SSI were both used during the course of the longitudinal study, but do not affect the identification criteria, as they measure stuttering severity using the same methodology in the non-readers age group.

Racial breakdown for the non-stuttering group was 3% American Indian/Alaska Native, 3% Black or African-American, 3% Hawaiian or Other Pacific Islander, 86% White, and 5% Multi-racial.

Exclusion criteria and attrition.—Of the 206 participants in the large-scale longitudinal study, 66 met the criteria for inclusion in the present study: 10 classified as *persisting*, 20 classified as *recovered*, and 36 classified as *non-stuttering*. Children were excluded from participating in the study if they scored below the 17th percentile on any of the speech and language tests and/or had a co-occurring developmental disability at any visit (n = 12 non-stuttering, 9 stuttering), or were non-compliant at the first visit (n = 2 non-stuttering, 2 stuttering). Reasons children did not complete the study, thereby not participating for at least 24 months as required in this study, included the family moving away (n = 4 non-stuttering, 1 stuttering), parents could not be contacted (n = 10 non-stuttering, 11 stuttering), parent request to withdraw (n = 17 non-stuttering, 15 stuttering) and undocumented reasons (n = 1 non-stuttering). These explanations contributed to an attrition rate of 28.6% for children who qualified but did not complete the study. Children who entered the study above the age of 4;11 were also excluded (n = 11 non-stuttering, 1 stuttering). Based on our group classification system, thirteen children were unclassifiable at the first visit, and four children who stuttered at the first visit were unclassifiable at their final visit due to differing results on the SSI and the 300-word disfluency count (e.g., a participant scored 11 or higher on the SSI, but exhibited below 3 stuttering-like disfluencies per 100 words in a 300-word conversational speech sample, or vice versa). Furthermore, ten children classified as non-stuttering at the first visit were also excluded due to an emergence of stuttering during the course of the longitudinal study. An additional 17 participants were excluded due to ECG recording issues.

Procedure

Caregivers and participants visited the university every 7–10 months for 22–32 months (based on availability). An examiner played with the participant while collecting a 300-word conversational speech sample. Following the speech sample, the participant completed a standardized test battery, which included the Peabody Picture Vocabulary Test-Fourth Edition (PPVT-4; L. M. Dunn & Dunn, 1997) to assess receptive vocabulary; the Expressive Vocabulary Test-Second Edition (EVT-2; Williams, 1997) to assess expressive vocabulary ability; Goldman-Fristoe Test of Articulation-Second Edition (GFTA-2: Goldman & Fristoe, 2000) as an articulation measure; and the Test of Early Language Development-Third Edition (TELD-3; Hresko, Reid, & Hammill, 1999), for both expressive and receptive language using the respective subscales. A pure-tone hearing test was also conducted to screen for hearing loss.

One week after speech and language testing, participants and their parents returned to the university. For this visit, participants were sat in a standard child car safety seat in front of a computer screen for data acquisition. Hypoallergenic adhesive electrodes were placed on the skin at the jugular notch and the base of the rib cage to acquire RSA. Participants viewed a 4-min screensaver of a fish-tank (initial baseline condition) and counterbalanced 4-min emotionally arousing positive and negative film clips from children's movies (e.g., *The Lion King*, *Little Mermaid*). A speaking task was performed following the initial neutral baseline video and after each of the positive and negative emotionally-arousing film clips. For the speaking task, participants were asked to produce a narrative ("tell a story") about text-less storybooks about a boy, a dog, and a frog by the author Mercer Mayer (e.g., *Frog Where Are You?*; Mayer, 1969). Three different books were used in the study, with the order counterbalanced across children.

As described by Jones et al. (2014), one PhD student and one Masters level research assistant were trained to code emotional expression in young children to validate the effectiveness of the audio/video clips in eliciting the expected positive, negative, and neutral emotions. The research personnel, blind to the experimental condition, viewed video recordings of 8 children who stutter and 8 children who did not stutter during the three experimental conditions (baseline, positive, negative) and predicted the experimental condition the children were viewing based solely on their behavior. The valence of the video (baseline, positive, negative) was accurately predicted for 79% of the viewing conditions, providing evidence that the experimental conditions elicited distinctive behavioral effects for each condition. This approach (behavioral observation) was used to examine emotional responses instead of self-reports due to the low reliability of emotional state reporting in young children.

Parasympathetic measure – Processing of respiratory sinus arrhythmia.—For this study, RSA at the initial time point was examined. To obtain RSA, the procedure outlined in Jones et al. (2014) was followed. Specifically, an electrocardiogram (ECG) was collected for each participant using the Biopac MP150 system (Biopac Systems, Inc.) and digitized at 1250 Hz. From these participants' extant data, the raw ECG was band-pass filtered to remove high frequency noise and low frequency drift (high pass cutoff: .5 Hz; low pass cutoff: 35 Hz). Biopac software was used to process the ECG signal by detecting the peak of the R-wave and timing the sequential inter-beat-intervals (IBI) in milliseconds. Inter-beat-interval time series were produced from segments within the ECG corresponding to each baseline, the negative and positive film clips, and the first 4 minutes of all three narrative tasks. Next, the IBI time series was processed by CardioEdit software (Brain-Body Center, University of Illinois at Chicago, 2007) to correct artifacts due to ventricular arrhythmias and faulty detections due to participant movement. For aberrant beat detections, consecutive points were added, and for missed beat detections consecutive points were divided to be consistent with surrounding points. No more than 5% of the total data for any condition was corrected using CardioEdit. CardioBatch software (Brain-Body Center, University of Illinois at Chicago, 2007) was used to derive measures of RSA from artifact-corrected IBI files. RSA was calculated using Porges' method (Porges, 1985; Porges & Bohrer, 1990), which uses a 21-point polynomial to detrend periodicities in the IBI series slower than RSA (e.g., basic metabolic processes often associated with vasomotor and blood pressure oscillation rather than RSA). A band-pass filter was then applied to the time-sampled IBI series (i.e., 250 msec intervals) to extract the variance at the frequency of respiration for young children (.24–1.04 Hz). Respiratory sinus arrhythmia (RSA) was expressed as the natural log of this variance: $\ln(\text{ms})^2$ (for further details, see Lewis et al., 2012). Values of RSA were derived for sequential 30-s epochs within each 4-min condition (i.e., baseline, positive and negative video viewing, and accompanying narrative speaking tasks,). The average of the 30-s epochs in each 4-min epoch was the metric for RSA in the data analyses." (p. 18–19.

Measurement reliability for RSA.—Twenty percent of the final data was randomly selected to calculate inter-rater reliability between research personnel for RSA. RSA values between rater pairs were within 0.10 ln $(ms)^2$ and correlations of RSA values between rater pairs were 0.99 for all conditions.

Data Analysis

The statistical models described below included fixed factors and continuous covariates that reflect alternative explanations for the observed results. Five variables were used as covariates for all statistical models: (1) sex, (2) race (3) age, (4) respiratory frequency, and (5) body mass index (BMI). Specifically, two fixed factors, sex and race, were covaried in each model. Age (in months) was included as a covariate since age is associated with RSA (El-Sheikh, 2005). Respiratory frequency was also entered as a continuous covariate since respiration has been shown to be associated with RSA (Hirsch & Bishop, 1981). Respiration rate was estimated from the frequency of RSA derived from the beat-to-beat heart rate pattern using a custom program, RespFreqFromRSA (Lewis, 2010). The frequency of RSA correlates with the frequency of respiration (correlation above .99), when respiration is measured via changes in chest circumference (see Denver et al., 2007). Body mass index was also included as a continuous covariate since some age groups have shown an association between body mass and RSA (Byrne et al., 1996; El-Sheikh, 2005). Children were measured for height and weight, with BMI computed using the standard height-weight index.

Statistical Modeling

Linear mixed-effects models (LMM, Pinheiro & Bates, 2000; Diggle et al., 2002) were employed to examine the three hypotheses. The LMM is similar to an ANOVA, but better controls for correlated errors occurring with repeated measurements of an individual (Nich & Carroll, 1997). Linear mixed-effects analyses were run using the MIXED package from Statistical Package for the Social Sciences Statistics version 24 (SPSS Statistics).

To address the *first hypothesis*, we ran a model to assess between-group differences in baseline RSA, measured during the neutral film clip, for the persistent, recovered, and non-stuttering groups. The between-group model includes the group factor and covariates of sex, race, age, respiratory frequency, and BMI.

To address the *second hypothesis*, we ran a model to assess between-group differences in RSA during the viewing of the positive video and the negative video for the persistent, recovered, and non-stuttering groups. The between-group model includes group, condition, and the interaction of Group × Condition to assess between-group differences in RSA with sex, race, age, respiratory frequency, BMI, and baseline RSA during the initial neutral video as covariates.

To address the *third hypothesis*, we ran a model to assess between-group differences in RSA during the narrative tasks following the viewing of the initial baseline neutral video, positive video, and negative video for the persistent, recovered, and non-stuttering groups. The between-group model includes group, condition, and the interaction of Group × Condition to assess between-group differences in RSA with sex, race, age, respiratory frequency, BMI, and baseline RSA during the initial neutral video as covariates.

Results

Descriptive Statistics

Table 1 includes participants' demographic, speech fluency, and speech/language characteristics at the initial visit. One-way analysis of variance models assessed between-group differences regarding age, SES, BMI, speech disfluencies, SSI scores, and standardized scores for the GFTA-2, PPVT-4, EVT-2, and the receptive and expressive subscales of the TELD-3. Chi-square analyses assessed between-group differences in sex and race. All descriptive statistical analyses were run using Statistical Package for the Social Sciences Statistics version 24 (SPSS Statistics).

Demographic Variables.—No between-group differences were found for chronological age in months for the persisting group, recovered group, and the non-stuttering group, $F(2,63) = 1.565$, $p = 0.217$. No between-group differences were found for race for the persisting group, recovered group, and non-stuttering group, $\chi^2(8) = 10.688$, $p = 0.220$. Similarly, no between-group differences were found for SES for the persisting group, recovered group, and non-stuttering group, $F(2,63) = 0.162$, $p = 0.851$. Between-group differences were found for sex with the persisting group (1 girl, 9 boys), and the recovered group (2 girls, 18 boys) significantly different from the non-stuttering group (20 girls, 16 boys), $\chi^2(2) = 14.957$, $p = 0.001$. Between-group differences were also found for BMI for the persisting group, recovered group, and non-stuttering group, $F(2,60) = 4.322$, $p = 0.018$. Post hoc follow-up comparisons revealed that the recovered group was significantly higher than the non-stuttering group (mean difference = 1.034, $p = .005$). As described in the Data Analysis section, both sex and BMI were included as covariates in the statistical analyses for the main hypotheses. See Table 1 for means and standard deviations.

Speech Fluency Variables.—In regard to stuttered disfluencies per 100 words, the persisting group (mean difference = 8.593, $p < .001$) and recovered group (mean difference = 5.923, $p < .001$) exhibited significantly more disfluencies than the non-stuttering group. The persisting group was also significantly higher than the recovered group (mean difference = 2.671, $p = .037$). In regard to SSI scores, the persisting group (mean difference = 12.271, $p < .001$) and recovered group (mean difference = 10.121, $p < .001$) exhibited significantly higher scores than the non-stuttering group. There were no significant differences in SSI scores between the persisting group and the recovered group (mean difference = 2.150, $p = .145$). See Table 1 for means and standard deviations.

Speech and Language Variables.—As seen in Table 1, there were no differences in GFTA-2, ($p = 0.229$), PPVT-4 ($p = 0.433$), TELD-3 receptive ($p = 0.196$), and TELD-3 expressive ($p = 0.631$). There were between-group differences in the EVT-2 ($p = 0.028$). Specifically, the non-stuttering group was significantly higher than the recovered group ($p =$ .010). Models of each study hypothesis with and without EVT-2 as a covariate were run, and statistical significance was unchanged. Findings of the analytical models without EVT-2 as a covariate are presented in the Results section.

Respiratory Sinus Arrhythmia and Childhood Stuttering

Comparing baseline RSA.—Inconsistent with the prediction that the persisting group would exhibit a lower capacity for emotion regulation indexed by lower baseline RSA, there were no significant differences in baseline RSA between the persisting, recovered, and non-stuttering groups, $F(2, 56.86) = 0.143$, $p = 0.867$, $d = .100$.

Comparing RSA during emotionally arousing video clips.: The *group × emotionally arousing video condition interaction* was significant, $F(2,698.00) = 11.079$, $p = < .001$, $d = .252$. Follow-up comparisons indicated the interaction was significant in the recovered and non-stuttering comparison, $F(1, 597.599) = 20.737$, $p < .001$. The recovered group exhibited lower RSA during the positive video clip and higher RSA during the negative clip (mean difference = −0.097, $p = .049$) while the non-stuttering group exhibited the opposite pattern with higher RSA during the positive video clip and lower RSA during the negative video clip (mean difference = 0.186, $p < .001$). Follow-up comparisons also indicated the interaction was significant in the persisting and the non-stuttering comparison, $F(1, 478.020) = 4.118$, $p = .043$. In contrast to the non-stuttering group, the persisting group did not exhibit significantly different RSA between the positive and negative video clips (mean difference = 0.020, $p = .776$). Follow-up comparison between the persisting group and the recovered group did not indicate a significant difference, $F(1, 320.868) = 0.471$, $p = .493$. See Figure 1 for results.

There was no significant main effect (viewing conditions combined) of group for RSA during the emotionally arousing video viewing conditions, $F(2, 79.02) = 0.147$, $p = .864$, d=.086. There was also no significant effect of condition, $F(1, 697.401) = 1.341$, $p = .247$, $d = .088$.

Comparing RSA during narratives following emotionally arousing video clips.: There was a significant effect of condition, $F(2, 975.52) = 43.266$, $p = < .001$, $d = .421$. Follow-up pairwise comparisons revealed that for all participants, RSA during the narrative following the neutral baseline video clip (estimated marginal mean [*EMM*] = 5.782, standard error (*SE*) = 0.189) was significantly greater than RSA during the narrative following the negative video clip (*EMM* = 5.418, SE = 0.189, $p < .001$, 95% C.I. = [−.459, −.270]), and during the narrative following the positive video clip (*EMM* = 5.375, *SE* = 0.190, $p < .001$, 95% C.I. = [−.501, −.313]). There was no significant group main effect of RSA during the narratives, $F(2, 136.50) = 0.095$, $p = 0.909$, $d = .053$. The *group × narrative condition interaction* was significant, $F(4, 974.97) = 3.028$, $p = 0.017$, $d = .111$. See Figure 2 for results. Follow-up comparisons indicated the interaction was significant in the persisting and non-stuttering comparison, $F(2, 638.437) = 4.858$, $p = .008$. The persisting group exhibited a greater decrease in RSA from the post-neutral to the post-positive narrative (mean difference = 0.612, $p < .001$) compared to the non-stuttering group (mean difference = 0.254, $p < .001$), and a greater decrease in RSA from the post-neutral to the post-negative narrative (mean difference = 0.524, $p < .001$) compared to the non-stuttering group (mean difference = 0.196, $p < .001$). Follow-up comparisons indicated that the interaction was not significant in the recovered group and persisting group comparison, $F(2, 447.894) = 2.117$, $p = 0.122$. In the recovered group, decreases in RSA from the post-neutral to post-positive narrative (mean difference = 0.355, $p < .001$) and the post-neutral to post-negative narrative (mean difference = 0.374, $p = .001$) did not significantly differ from previously mentioned decreases in the persisting group or the non-stuttering group. Follow-up comparisons also indicated the interaction effect was not significant in the recovered group and non-stuttering group comparison, $F(2, 840.895) = 1.814$, $p = 0.164$.

Follow-up Analyses—We conducted two follow-up analyses. These follow-up analyses were motivated by our finding that persisting children exhibited higher RSA during the narrative following the neutral video (relative to the positive and negative conditions) compared to the nonstuttering group, and were designed to provide insight on the possibility that children who stutter may exhibit relatively slower changes in cardiac activity during social communicative situations (Jones, Buhr, et al., 2014).

RSA during the baseline (neutral) and post-neutral speaking condition.: Our first follow-up analysis was designed to determine whether there were differences in parasympathetic activity during the baseline (neutral) viewing condition compared to the post-neutral speaking condition. This analysis specifically tests whether children who stutter and persist exhibit differences in parasympathetic cardiac activity from a neutral viewing condition to a speaking condition that immediately followed. There was a significant effect of condition, $F(1, 656.50) = 71.660$, $p = < .001$. Follow-up pairwise comparisons revealed that for all participants, RSA during the baseline (neutral) viewing condition was significantly greater than RSA during the post-neutral narrative (mean difference = 0.325, $p < .001$). There was no significant group main effect, $F(2, 90.180) = 0.464$, $p = 0.630$. The *group × narrative condition interaction* was significant, $F(2, 659.764) = 11.742$, $p < 0.001$. Follow-up comparisons indicated that the recovered group (mean difference = 0.422, $p < .001$) and non-stuttering group (mean difference = 0.516, $p < .001$) exhibited significantly greater RSA during the baseline (neutral) viewing condition compared to the post-neutral speaking condition, but the persisting group (mean difference = 0.038, $p = 0.666$) did not exhibit a difference in RSA between the two conditions.

Association between heart period and RSA during narratives.: Our second follow-up analysis assessed the association between heart period and RSA during the narratives, which was designed to provide insight on the vagal brake efficiency of the heart in response to activity that triggers autonomic reactivity (Dale et al., 2022; Kolacz et al., 2021). Greater association between heart period and RSA indicates stronger influence of parasympathetic activity (i.e., “cardiac vagal tone”) on overall cardiac output (heart period) during challenge (i.e., the amount of cardiac change associated with changes in RSA).

The specific follow-up analyses involved a linear mixed-effects model that was constructed to assess between-group differences in the association between heart period and RSA during the 30-sec epochs across each of the narrative speaking tasks. Heart period was the dependent variable in the model. The between-group model included RSA, group, condition, and the interaction of group × RSA to assess between group differences in the association between RSA and heart period. Further, sex, race, age, respiratory frequency, and BMI were included as covariates.

There was a significant effect of RSA, $F(1, 1285.94) = 479.26$, $p < .001$, which indicated a significant positive association between RSA and heart period for all participants. Further, the *group × RSA interaction* was significant, $F(2, 1257.97) = 12.830$, $p < .001$. The persisting group ($\beta = -10.37$, SE = 2.43, $p < .001$) and recovered group ($\beta = -6.58$, SE = 1.75, $p < .001$) exhibited a significantly less positive association between RSA and heart period compared to the nonstuttering group. There was no significant difference in the association between RSA and heart period between the persisting and recovered groups ($\beta = 3.80$, SE = 2.58, $p = .142$).

Summary of Main Findings—The present study resulted in several findings. First, the persisting group, recovered group, and non-stuttering group did not significantly differ in baseline RSA as predicted (Hypothesis 1). Second, during the emotionally arousing video clips, there was a significant *group × condition* interaction with the recovered group and the persisting group differing from the non-stuttering group in their responses to the positive and negative videos (Hypothesis 2); specifically, 1) the recovered group demonstrated higher RSA during the negative video clip and lower RSA during the positive clip and 2) the persisting group did not demonstrate significantly different RSA during the positive and negative video clips. Third, during the narrative tasks following the video clips, all three groups exhibited higher RSA during the speaking task following the neutral video with significantly lower RSA during the speaking tasks following the positive and negative videos, a level of RSA response that was greater for the persisting than the non-stuttering group (Hypothesis 3). Follow-up analyses indicated that: 1) the recovered and non-stuttering groups, compared to the persisting group, exhibited significantly greater RSA during the baseline (neutral) viewing condition compared to the post-neutral speaking condition, and 2) the non-stuttering group exhibited a significantly more positive association between heart period (overall cardiac output) and RSA (parasympathetic activity) during the narrative tasks than the recovered and persistent groups.

Discussion

The main findings will be discussed in terms of the three hypotheses, which will be followed by a discussion of caveats, limitations, and future directions.

Baseline RSA

Our first hypothesis predicted that the persisting group would exhibit a lower capacity for emotion regulation, indexed by significantly lower baseline RSA, compared to the recovered group and the non-stuttering group. Present results comparing baseline RSA between the groups did not support this prediction. This hypothesis was based on theoretical speculation (Walden et al., 2012) and empirical evidence (Ambrose et al., 2015; Erdemir et al., 2018) that aspects of emotion may contribute to stuttering persistence. To the extent that baseline

RSA represents a stable physiological measure of emotion regulation capacity (Butler et al., 2006; Demaree et al., 2004; Hastings et al., 2008), these results appear to be congruent with Ambrose et al.'s (2015) findings that effortful control did not differ between recovered, persistent and nonstuttering children. However, in a study of self-regulation at a much earlier age, Koenraads et al. (2021) found that children who later persisted were less able to "recover from distress" at 6 months of age compared to those that went on to recover. These researchers also noted that their results explained a small proportion of the variation in their study. Thus, while it is possible that temperamentally-related aspects of emotion regulation play a role in stuttering persistence, it is also quite possible, as speculated by Koenraads et al. (2021), that other variables are more influential than emotion regulation (see Singer et al., 2020 for review of factors associated with persistence). An additional consideration is that findings to date that have shown a significant association between stuttering persistence and emotional processes have largely implicated emotional reactivity (Ambrose et al., 2015; Erdemir et al., 2018; Koenraads et al., 2021; Singer et al., 2019; Zengin-Bolatkale et al., 2018). Therefore, while past cross-sectional research has implicated regulation in developmental stuttering in this age range (e.g., Anderson et al., 2003; Eggers et al., 2010; Karrass et al., 2006), the current evidence provides more support for a potential role of reactivity in stuttering persistence. Future studies with larger sample sizes are clearly necessary to establish a comprehensive understanding of the role of emotional processes in stuttering persistence. These studies may consider using diverse methodologies as well as investigating the interplay between reactivity and regulation processes.

Our baseline RSA hypothesis was also based on previous empirical evidence indicating that young children who stutter exhibited lower levels of baseline RSA than their non-stuttering peers (Jones, Buhr, et al., 2014). This present study and our previous work were comprised of partly overlapping samples (24 participants were included in both Jones et al. [2014] and the current study, representing 36% of the current study sample), identical subject recruitment methods, and data collection procedures. It should be noted that the two studies employed different group classification procedures, therefore, the results are not directly comparable. The current study examined persisting, recovered, and non-stuttering groups whereas Jones et al., (2014) consisted only of stuttering and non-stuttering groups. Despite this, we had still expected that children who stutter and recover and/or children who stutter and persist to exhibit lower levels of baseline RSA than the non-stuttering children given that they were both classified as children who stutter (CWS) at timepoint 1 in this study. However, given the high reliability between RSA coders for this study and the past study, we believe that the lack of effect for baseline RSA is due to sampling error and differential levels of baseline RSA in the non-overlapping participants in the previous study compared to the current study (for an example of a study when previous cross-sectional findings were not replicated by the same lab in a subsequent longitudinal study, see Hilger et al., 2016). Further, given that the previous results (Jones, Buhr, et al., 2014) just reached the level of statistical significance ($p = .04$), the present results underscore the need for larger samples to critically evaluate whether baseline levels of RSA are indeed associated with stuttering in young children.

RSA During Emotionally Arousing Videos

We hypothesized that while watching emotionally arousing videos, the persisting group would exhibit lower emotion regulation, indexed by a higher level of RSA adjusted for baseline, compared to the recovered group and the non-stuttering group. This speculation was based on theoretical models suggesting that children who stutter, compared to children who do not stutter, show decreased emotion regulation in emotionally arousing situations (Anderson & Felsenfeld, 2003; Johnson et al., 2010; Jones et al., 2014; Ntourou et al., 2013; Walden et al., 2012). Present results, with no significant main effects of group or condition, do not support this speculation for emotion regulation across emotionally arousing conditions. However, the significant *group × condition interaction* shows different patterns of RSA response for the recovered group and non-stuttering group. The recovered group exhibited significantly lower RSA in the positive condition than in the negative condition whereas the non-stuttering group showed the opposite pattern, with higher RSA in the positive condition than the negative condition. These results suggest that greater reactivity to positive conditions (relative to negative) may be associated with transient stuttering, or short-term stuttering followed by recovery, which was not the case for children with persisting stuttering. This finding is supportive of past research that has also found that children who stutter exhibited increased stuttering during positive conditions (Choi et al., 2016; Johnson et al., 2010) and highlights the possibility that emotional arousal related to various contexts and conditions may be related to stuttering. In this case, the children who stutter and recover, a subset of the children who stutter, may be more reactive to positive emotions and, thus, increase emotional regulation. This increased regulation during positive emotion conditions is consistent with literature related to capacity to regulate the vagal brake, where greater attentive involvement or mental effort is associated with greater reduction in RSA (Walter & Porges, 1976), and may ultimately be a process associated with the trajectory of stuttering.

RSA During Narratives Following Emotionally Arousing Videos

We hypothesized that during the speaking tasks following each of the video clips, the persisting group would exhibit more limited emotion regulation—a less effective vagal brake (Porges et al., 1996) indexed by a higher level of RSA adjusted for baseline, compared to the recovered group and the non-stuttering group. This hypothesis was supported by the significant *group × narrative condition interaction*, which indicated that the persisting group exhibited higher RSA during the neutral narrative (relative to the negative and positive narratives) compared to the non-stuttering group. There were no differences between the persisting and recovered groups. However, follow-up analyses indicated that both the recovered and the nonstuttering groups, compared to the persisting group, exhibited significantly greater RSA during the baseline (neutral) viewing condition compared to the post-neutral narrative condition. One potential implication of these results is that the persisting group's increased RSA during the neutral narrative (relative to the non-stuttering group) and lack of difference between the baseline viewing and post-neutral speaking condition (compared to the recovered and nonstuttering groups) may reflect a slowness to release the vagal "brake" and engage in the novel speaking task. The inefficiency of vagal brake release during challenge has been described in children with selective mutism, who struggle initiating speech (Heilman et al., 2012). For children who stutter, the higher

RSA observed in the narrative following the neutral video clip and the lack of decrease in RSA from the baseline condition may be consistent with having a sluggish vagal brake, which may in turn impact efficient engagement in social-communication situations. This speculation is in line with our previous cross-sectional findings that children who stutter, compared to children who do not stutter, exhibit higher RSA during speaking conditions (Jones, Buhr, et al., 2014). It is also consistent with findings by Choi et al. (2013) indicating that children who stutter more often exhibited behavioral inhibition in the "extremely high" (top 15% of the distribution) range compared to children who do not stutter. Therefore, it is possible that higher RSA during the post-neutral narrative for the persisting group indicates higher behavioral inhibition and/or a slowness to engage during a novel speaking task that is not present in subsequent speaking tasks after the participant has had the opportunity to acclimate to the experimental conditions. An additional consideration is that lower RSA (and more pronounced change from the baseline condition to the post-neutral speaking condition) for the recovered and non-stuttering groups may depict a more adaptive response to the neutral narrative task, similar to studies demonstrating that children who display lower levels of RSA during emotionally and cognitively challenging narrative tasks exhibit the most coherent and adaptive narratives (Bar-Haim et al., 2004).

Another potential interpretation of the current results on RSA during speaking is that the persisting group exhibits a more pronounced change in RSA from the post-neutral narrative to the post-positive and post-negative narratives. As mentioned earlier, moderate, as opposed to large, decreases in RSA are likely more adaptive and facilitate cognitive and attentional processes, both of which are required for effective performance during a narrative speaking task. Therefore, the persisting group's greater difference from one condition to the next may be less adaptive during emotionally arousing speaking conditions. This speculation is consistent with our findings that larger decreases in RSA from baseline are associated with increased stuttering during speaking tasks (Jones et al., 2017), although it should be noted that we did not directly assess the association between stuttering frequency and changes in RSA in the current study.

Lastly, our follow-up analyses also indicated a significantly more positive association between RSA and heart period for the nonstuttering compared to persisting and recovered groups. A potential interpretation of this finding is that there is a lesser influence of parasympathetic activity on overall cardiac output for both the persistent and recovered groups, compared to the non-stuttering group, during social-communicative challenges. In essence, for children who stutter, the vagal brake may be less efficient at mobilizing cardiac activity during situations when relative quick autonomic responses are likely to be adaptive. This finding provided support for the aforementioned speculation that children who stutter may be characterized by a "sluggish vagal brake" or overall slowness to engage as discussed above. Further study, specifically designed to test for the efficiency of vagal responses and cardiac output across a variety of conditions that trigger autonomic reactivity (e.g., Dale et al., 2022; Kolacz et al., 2021; Porges et al., 2019), is necessary to provide further insight on the potential that children who stutter exhibit less vagal efficiency.

Caveats and limitations

The present study attempted to account for the possible effect of talking duration on respiration and RSA. However, the best approach for accounting for potential influence of speech-related respiration on RSA remains unclear. Further studies are needed to determine if speaking (and other activity) impacts the quantification or interpretation of RSA. This study also assessed RSA during a speaking task but did not assess the association between instances of stuttering and RSA; considering the innervation of the pharynx and the larynx by the pharyngeal and superior laryngeal branches of the vagus nerve, this question awaits future empirical study. Additionally, since study participants had no known speech or language disorders other than stuttering, with typical hearing and typical neurological, intellectual, and social development, present findings may more directly reflect a subgroup of children who stutter without the aforementioned deficits. Lastly, the present study had a relatively small sample size, especially for the group of children who stutter and persist. This decreases the statistical power and increases the possibility of Type II error.

Future directions

Future studies may further examine longitudinal changes in emotion regulation via changes in baseline RSA and task-dependent RSA between the persisting, recovered, and non-stuttering groups at each longitudinal time point. We could then examine how changes in capacity and fluidity to regulate emotions, as indexed as RSA, may differ between the groups throughout the process of group determination. For example, does the persisting group continue to exhibit similar baseline RSA to the recovered and persisting groups at the final time point, or does the persisting group exhibit a changing trajectory? Does the recovered group continue to exhibit increased reactivity to positive emotionally-arousing conditions at the final time point, or is this response diminished as disfluency decreases?

Conclusion

The study's main hypotheses were partially supported. There were nuanced findings (group × condition interactions) that differentiated the recovered and persisting groups from the nonstuttering group in our main analyses. For instance, there were differences in response to the positive and negative video clips, with the recovered group exhibiting increased regulation, indexed by decreased RSA, in the positive condition (relative to the negative). Further, compared to the non-stuttering group, the persisting group exhibited larger differences in RSA during the post-positive and post-negative narrative conditions compared to the post-neutral narrative condition. Lastly, a follow-up analysis indicated that the recovered and nonstuttering groups, compared to the persisting group, exhibited significantly greater RSA during the baseline (neutral) condition compared to the post-neutral narrative task. These findings provide a physiological perspective of emotion regulation to augment earlier investigations of stuttering persistence that demonstrated that children who stutter and persist, compared to those who recover, exhibited heightened negative affect (Ambrose et al., 2015) and increased physiological reactivity (Zengin-Bolatkale et al., 2018). Future investigations with larger sample sizes and diverse methodologies are necessary to provide novel insights on the specific emotion-related processes that are potentially involved with persistence of stuttering in young children.

Acknowledgements

This work was supported by National Institutes of Health (NIH) grants from the National Institute on Deafness and Other Communication Disorders (NIDCD) to Vanderbilt University (5R01DC000523-19, 2R56DC000523-20A1) and Vanderbilt University Medical Center (R21DC016723, R01DC020311), as well as Vanderbilt CTSA grants from NCATS/NIH (UL1RR024975, UL1TR00044506, UL1TR002243), and a Vanderbilt Kennedy Center Hobbs Discovery Grant. This research was also supported by the Wilker-Ellis Stuttering Research Fund at Vanderbilt University Medical Center. The content reported herein is solely the responsibility of the authors and does not necessarily represent the official views of the NIH, NIDCD, Vanderbilt University, Vanderbilt University Medical Center, the Vanderbilt Kennedy Center, or the generous donors that supported this work. The authors also like to sincerely thank the children and caregivers of this study—this project would not have been possible without their participation.

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Highlights

- Temperament and emotion have both been implicated in stuttering.
- Respiratory sinus arrhythmia (RSA) indexes emotion regulation.
- RSA may be associated with stuttering persistence or recovery.
- Group × condition interactions differentiated the persisting stuttering group.

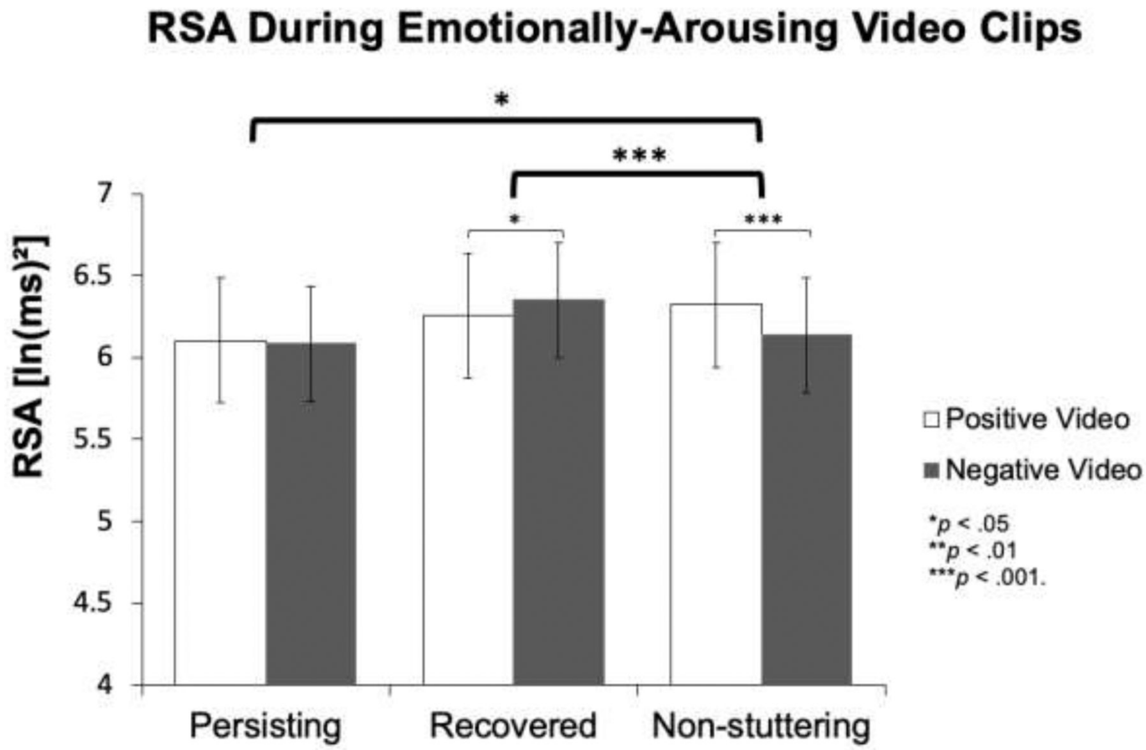

Figure 1.

RSA values during emotionally-arousing video clips. Error bars represent standard error. Bold brackets indicate a significant follow-up comparison for the *group × video condition* interaction. *$p < .05$, **$p < .01$, ***$p < .001$.

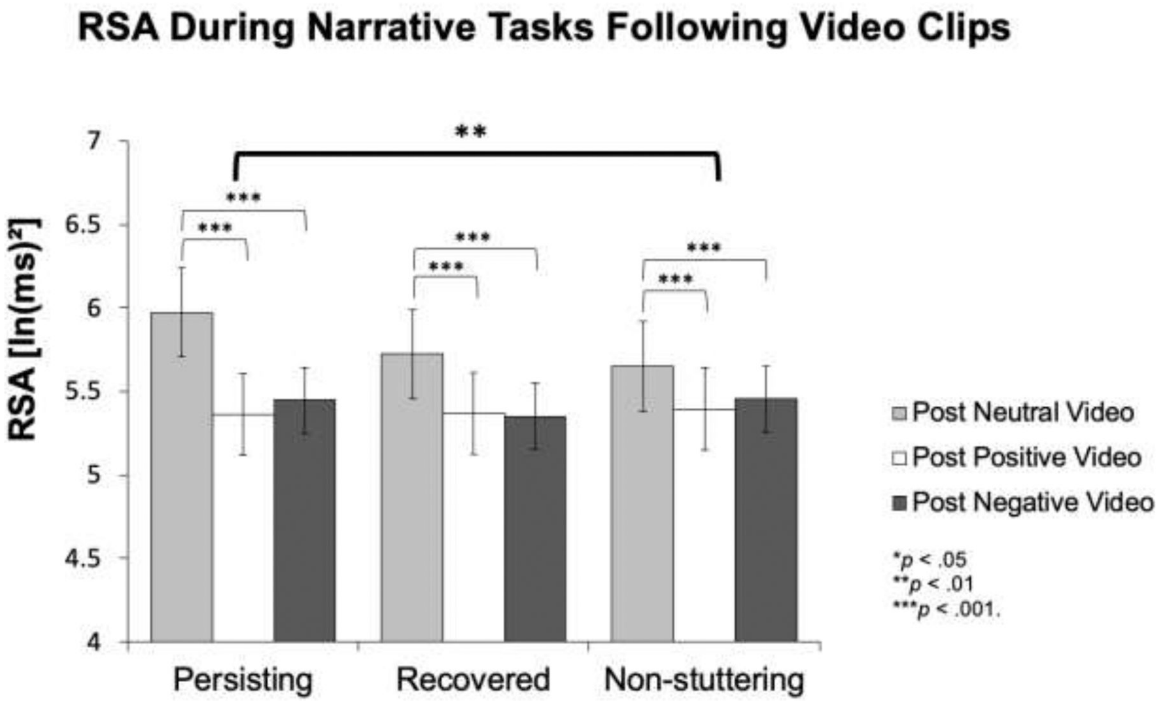

Figure 2.

RSA values during narrative tasks following neutral, positive, and negative video clips. Error bars represent standard error. Small brackets indicate significant pairwise follow-up comparisons for the *condition effect*. The bold bracket indicates a significant follow-up comparison for the *group × narrative condition* interaction. *$p < .05$, **$p < .01$, ***$p < .001$.

Table 1.

Demographic, speech fluency, and speech/language characteristics of the persisting group, recovered group, and the non-stuttering group.

Variable	Persisting M (SD)	Recovered M (SD)	Non-stuttering M (SD)	*F* (df)	Wald X^2(df)	*p*	Effect size
Demographic characteristics							
Chronological age (mos.)	46.6 (4.5)	44.7(6.0)	47.8 (6.8)	1.565 (2, 63)		0.217	.315
Gender					14.957 (2)	0.001	
Race					10.688 (8)	0.220	
SES	45.1(10.5)	45.5 (10.7)	46.9 (11.6)	0.162 (2, 63)		0.851	.101
BMI	15.8 (1.3)	16.7 (1.6)	15.7 (1.0)	4.322 (2, 60)		0.018	.537
Speech Fluency Measures							
Stuttering Frequency (%)	9.9 (6.5)	7.3 (3.6)	1.3 (0.7)	37.926 (2, 62)		<.001	1.564
SSI-3 total score	19.3 (6.8)	17.2 (4.6)	7.03 (1.3)	68.269 (2, 62)		<.001	2.099
Speech/Language Testing							
EVT	117.4 (10.8)	115.4 (10.2)	122.8 (9.8)	3.802 (2, 63)		0.028	.491
PPVT	121.1 (14.7)	116.5 (12.3)	120.3 (10.3)	0.848 (2, 63)		0.433	.232
GFTA	107.6 (10.6)	110.7 (7.7)	112.8 (8.5)	1.507 (2, 63)		0.229	.309
TELD Receptive	115.6 (13.4)	124.6 (14.2)	122.0 (12.9)	1.673 (2, 63)		0.196	.325
TELD Expressive	110.9 (16.7)	114.0 (19.7)	116.0 (11.8)	0.464 (2, 63)		0.631	.171