HHS Public Access
Author manuscript
J Fluency Disord. Author manuscript; available in PMC 2019 June 01.

Published in final edited form as:
J Fluency Disord. 2018 June ; 56: 1–17. doi:10.1016/j.jfludis.2017.11.003.

The effect of emotion on articulation rate in persistence and recovery of childhood stuttering

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Abstract

Purpose—This study investigated the possible association of emotional processes and articulation rate in pre-school age children who stutter and persist (persisting), children who stutter and recover (recovered) and children who do not stutter (nonstuttering).

Methods—The participants were ten persisting, ten recovered, and ten nonstuttering children between the ages of 3 to 5 years; who were classified as persisting, recovered, or nonstuttering approximately 2–2.5 years after the experimental testing took place. The children were exposed to three emotionally-arousing video clips (baseline, positive and negative) and produced a narrative based on a text-free storybook following each video clip. From the audio-recordings of these narratives, individual utterances were transcribed and articulation rates were calculated.

Results—Results indicated that persisting children exhibited significantly slower articulation rates following the negative emotion condition, unlike recovered and nonstuttering children whose articulation rates were not affected by either of the two emotion-inducing conditions. Moreover, all stuttering children displayed faster rates during fluent compared to stuttered speech; however, the recovered children were significantly faster than the persisting children during fluent speech.

Conclusion—Negative emotion plays a detrimental role on the speech-motor control processes of children who persist, whereas children who eventually recover seem to exhibit a relatively more

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stable and mature speech-motor system. This suggests that complex interactions between speech-motor and emotional processes are at play in stuttering recovery and persistency; and articulation rates following negative emotion or during stuttered versus fluent speech might be considered as potential factors to prospectively predict persistence and recovery from stuttering.

1. Introduction

Stuttering is a developmental disorder with an average age at onset of 30–36 months, and a lifetime incidence of 5–8% (Yairi & Ambrose, 2013). Approximately 75–85% of children who stutter recover from the disorder before age 7 with little or no professional treatment (Yairi & Ambrose, 1999, 2005, 2013), however, the remaining 15–25% continue to stutter into adulthood (Yairi & Ambrose, 1999). For those affected by the disorder, stuttering can have a significant and life-long negative impact on social (Van Borsel, Brepoels, & De Coene, 2011), educational (O'Brian, Jones, Packman, Menzies, & Onslow, 2011), and vocational development (Klein & Hood, 2004), as well as emotional well-being (Tran, Blumgart, & Craig, 2011; Treon, Dempster, & Blaesing, 2006). Also, stuttering is associated with negative experiences such as bullying (Blood & Blood, 2007). Thus, it is imperative to develop a better understanding of factors contributing to the development of stuttering.

Despite an extensive body of literature investigating the mechanisms contributing to stuttering (for reviews, see Bloodstein & Bernstein Ratner, 2008; Conture & Walden; 2012; Yairi & Ambrose, 2005), the nature of the disorder is still poorly understood. Many scholars describe childhood stuttering as a complex and multifaceted disorder, and argue that a unidimensional characterization is not possible. At present, studies of the mechanisms contributing to recovery and persistence of stuttering are limited (Reilly et al., 2009; Yairi & Ambrose, 2005; 2013). Generally acknowledged, however, is that multiple factors contribute to persistence versus recovery from stuttering, including linguistic and phonological factors (Mohan & Weber, 2015; Watkins & Yairi, 1997; Yairi & Ambrose, 2005; Usler & Weber-Fox, 2015), speech-motor factors, (Spencer & Weber-Fox, 2014; Usler, Smith, & Weber, 2017), physiological/anatomical factors (Chang, Erickson, Ambrose, Hasegawa-Johnson, & Ludlow, 2008; Chang, Zhu, Choo, & Angstadt, 2015; Chang, Angstadt, Chow, Etchell, Garnett, Choo, Kessler, Welsh, & Sripada, 2017), cognitive factors (Chang et al., 2017), and temperamental/emotional factors (Ambrose, Yairi, Loucks, & Seery, 2015).

1.1 Stuttering, Recovery and Persistence

Factors that contribute to the onset of stuttering and possibly predict recovery and persistence are best detected before extensive experience with stuttering leads to its own impact on speech-motor execution, linguistic, cognitive, and emotional processes. This warrants increased longitudinal study of preschool-age children who are close to the onset of stuttering to disentangle factors that contribute to persistent stuttering from consequences of experience with stuttering. Also, treating children who stutter (CWS) as a single group, which often has been done in previous cross-sectional studies, might mask potential factors that lead to recovery/persistence, and may be one reason why findings are often mixed. Moreover, being able to predict persistency/recovery prospectively is important from a clinical perspective (Watkins & Yairi, 1997; Yairi, Ambrose, Paden, & Throneburg, 1996), since it would enhance our ability to identify CWS at high risk to persist and promote the continued development of early intervention tools for children at high-risk. Therefore, there is a clear need to identify factors that initiate, discontinue and perpetuate stuttering in young children.

Stuttering is characterized by disruption of speech fluency, which reflects a breakdown in the speech-motor system. Fluent speech requires accurately timed vocal movements that depend on a well-functioning speech-motor execution system. Therefore, speech-motor processes may play a role in persistent-recovery from childhood stuttering. In line with this, Spencer and Weber-Fox (2014) reported that persisting children compared to recovered children demonstrated poorer performance at their initial visit (which predicted eventual recovery or persistence) in speech articulation and novel non-word repetition tasks, both involving speech-motor programming and execution processes. In another longitudinal study of children aged 5;9–8;0 Usler et al. (2017) found that when compared with children who recovered, children with persistent stuttering exhibited higher lip aperture variability during production of sentences with varying lengths and syntactic complexities, indicating less refined and less mature articulatory speech-motor coordination. However, at the time of testing the children were already classified as persisting or previously recovered from stuttering. Ambrose et al. (2015), in a longitudinal study of 2–4 year-old children attempted to discover differences before the children were categorized into one of the two subgroups and reported greater speech kinematics variability as measured by jaw displacement and fundamental frequency variability in persisting than recovered children. However, the difference was documented only during the final visit, 2 years after the first visit, not at the first visit when children were within twelve months of the reported onset of stuttering.

Another key factor differentiating persisting and recovered stuttering could be temperament or emotion, as there is considerable evidence linking temperamental characteristics and emotional processes to stuttering in preschool-age children (see reviews by Conture, Kelly, & Walden, 2013; Jones, Choi, Conture, & Walden, 2014; Kefalianos, Onslow, Block, Menzies, & Reilly, 2012; cf. Alm, 2014). For example, a longitudinal study by Ambrose et al. (2015) showed that mothers of persistent children rated their children as having higher “negative affectivity” than mothers of recovered or control children. The children were 2–4 years of age and within 12 months of the reported onset of stuttering. Two components of the Negative Affectivity subscale of the Child Behavior Questionnare (Rothbart, Ahadi, Hershey, & Fisher, 2001) differentiated the groups: fear and soothability. Persisting children were rated as more fearful and less soothable by their mothers. Further, a recent study by Chang et al. (2017) in which resting state fMRI scans were collected from children between 3;3–10;8 years of age found anomalous connectivity among brain networks that support attention, motor processes, perception and emotion, which predicted both stuttering status and stuttering persistence.

Articulation rate has long been considered to be a marker of the speech-motor execution system, as it depends on temporal coordination of respiratory, articulatory and phonatory processes (Hall, Amir, & Yairi, 1999; McClean & Tasko, 2003; Tasko, McClean, & Runyan, 2007; Tumanova, Zebrowski, Throneburg, & Kayikci, 2011). However, we propose that articulation rate reflects a combination of speech-motor execution and emotional processes (for the link between speech rate and emotion see Bachorowski & Owren, 1995; Johnstone & Scherer, 2000; Scherer, 2003), and could be an early marker to differentiate persistency from recovery. A few studies have investigated articulation rate of preschool-age children who are later categorized as exhibiting persistent or recovered stuttering (Hall et al., 1999; Kloth, Kraaimaat, Janssen, & Brutten, 1999); but to our knowledge no study has investigated articulation rate in relation to emotion in this population. Speech rate is known to depend on the emotional status of the talker (e.g., we talk faster when we are excited); however, we do not know whether emotion-related changes in speech rate differentiate recovered from persisting children. Since articulation rate reflects a combination of speech-motor execution and emotional processes, it could be associated with recovery or persistence of stuttering in young children.

1.2 Articulation Rate and Stuttering

Articulation rate has been studied in people who stutter (PWS); however, there are few studies with young children, especially studies contrasting persisting and recovered. Moreover, findings have been mixed and inconclusive (for a review see Sawyer, Chon, & Ambrose, 2008). Several factors could account for discrepancies, including differences in: (1) research methods (e.g., whether pauses and disfluencies are removed while measuring articulation rate) (2) the metric of the measure used (syllables, words, or phones per second or minute), (3) types of utterances used for analysis (e.g., whether only perceptually fluent utterances are used), (4) heterogeneity of the samples in terms of age, time since onset of stuttering, and stuttering severity, (5) sample selection and (6) sample sizes.

Articulation rate measured on the fluent portion of speech can be thought of as an accurate measure of speech rate when the speech includes disfluencies. However, in the past, speech rate traditionally has included pauses and disfluencies (Bloodstein, 1944; Walker, 1988). This approach biases the rate of speech for PWS because stuttering includes disfluencies that slow down speech. Therefore, articulation rate as expressed by the number of syllables or phones per second after excluding pauses and disfluent segments is the optimal estimate of speech motor execution rate.

There are two major hypotheses about the link between stuttering and speech rate. According to one, children who stutter (CWS) speak slower, because they require additional time for linguistic and phonological planning of speech-motor actions, which results in delay in the speech-motor output (Perkins, Kent & Curlee, 1991; Peters, Hulstijn & Starkweather, 1989; Postma & Kolk, 1993). Supporting that hypothesis, Meyers and Freeman (1985) found that preschool-age CWS, especially those with more severe stuttering, spoke at slower rates during fluent speech than peers who did not stutter (CWNS). Conture, Louko, and Edwards (1993), on the other hand, hypothesized that CWS might speak at a rate that exceeds their speech-motor control capacity and/or language abilities, increasing the chances of disfluency. This perspective is reflected in advice given to CWS and their parents to reduce their speech rate to improve speech fluency. In line with this, Kloth, Janssen, Kraaimaat, and Brutten (1995), who tested young children at risk for developing stuttering due to family history, found that children who later stuttered had faster articulatory rates during perceptually fluent utterances than those who did not develop stuttering. Similarly, Kelly and

Conture (1992) found that CWS had faster articulation than CWNS during fluent utterances. The majority of studies, however, report no significant differences in articulation rate between CWS and CWNS (Chon, Ko, & Shin, 2004; Kelly & Conture, 1992; Logan, Byrd, Mazzocchi, & Gillam, 2011; Ryan, 1992, 2000).

Only two longitudinal studies, to our knowledge, examined the possibility that speech rate predicts persistence or recovery from stuttering. Hall et al., (1999), studied perceptually fluent utterances of children who later recovered from stuttering and those whose stuttering persisted and found that recovered children spoke at a slower rate near onset of stuttering than fluent peers but found no difference between persisting and recovered children. Kloth et al. (1999) examined fluent utterances of children before stuttering onset and one year after onset and reported that persisting children had higher variability in articulation rate than recovered children at both time points. Persisting children also appeared to have higher post-onset articulation rates than recovered children, however, the difference was not significant ($p = .09$).

Tumanova et al. (2011) found a negative correlation between articulation rate (measured on fluent portions of speech only) and frequency of stuttering-like disfluencies, showing that CWS with more stuttered disfluencies also had slower articulation rates. Using a similar method for articulation rate, Chon, Sawyer and Ambrose (2012) investigated whether articulation rate differed between several types of utterances in preschool age CWS: perceptually fluent (FLU utterance), containing normal disfluencies (OD utterance), containing stuttering-like disfluencies (SLD utterance), and containing both normal and stuttering-like disfluencies (SLD+OD utterance). However, there was no significant difference in the articulation rates between the fluent utterances and those with disfluencies that are stuttering-like, nonstuttering-like or both, despite the prediction that speech-motor control would act differently for temporal coordination of speech segments in fluent and disfluent speech. Their motivation was based on empirical reports (Yaruss & Conture, 1996; Logan & Conture, 1995) indicating slower articulation rates in stuttered than perceptually fluent utterances, despite the differences *not* being statistically significant. Accordingly, it has been reported that CWS with a relatively higher frequency of stuttering-like disfluencies (SLDs) have overall slower articulation rates, as there are anticipatory, carry-over or overflow effects (Zebrowski, 1994) in fluent segments preceding or following stuttered segments in the utterances (Tumanova et al., 2011). In line with this reasoning fluent speech surrounding instances of stuttering-like disfluencies might be slower than fluent speech that is not in the area of SLDs.

In sum, the mixed picture presented by previous studies suggest that articulation rate, in and of itself, may not provide a reliable variable for distinguishing persistence and recovery of stuttering. However, considering articulation rate in relation to emotional processes, rather than treating it exclusively as a measure of speech-motor execution, might provide additional insights into potential differences between these two sub-populations of CWS.

1.3 Articulation Rate and Emotionality

Acoustic properties of speech provide information about emotions. For example, the underlying emotional state of a speaker affects fundamental frequency, amplitude, rate and so forth (Bachorowski & Owren, 1995). Traditionally, researchers have experimentally studied acoustic parameters of emotion in adults by either inducing specific emotional states (Bachorowski & Owren, 1995; Karlsson, Beanziger, Dankovicova, Johnstone, Lindberg, Melin, Nolan, & Scherer, 1998; Scherer, 1986), or by asking actors to produce speech to convey specific emotions (Banse & Scherer, 1996; Scherer, Koivumaki, & Rosenthal, 1972; Scherer, London, & Wolf, 1973; Sobin & Alpert, 1999; Whiteside, 1999). It has been argued that the most fundamental determinants of such vocal changes are physiological changes in the body, such as responses in the autonomic nervous system associated with different emotions (see the reviews by Johnstone & Scherer, 2000; Scherer, 1986; Scherer, 2003). Given that different emotions are associated with different patterns of ANS activity (Ekman, Levenson, & Friesen, 1983; Levenson, 2003, 2014) these studies theorized a possible link between emotional states and vocal changes via physiology. That is, emotion specific activity in the sympathetic and parasympathetic pathways may lead to changes in the functioning of the speech production system and processes related to respiration, vocal fold vibration, phonation and articulation. Thus, there would be correspondence between specific acoustic parameters and underlying emotional states.

For example, negative emotions such as fear or anger have been linked to high sympathetic arousal, generating a fight-or-flight response that leads to increased blood flow to muscles, increased heart rate and muscle tension to provide the body with extra energy, speed and strength (Johnstone & Scherer, 2000; Scherer, 2003). Such physiological response, then, would be accompanied by an overall increase in rate of articulation (Ellgring & Scherer 1996; Johnstone & Scherer, 2000; Scherer, 2003; Schröder, 2001). Similarly, positive emotions, such as joy or happiness, have been linked to increases in articulation rate (Johnstone & Scherer, 2000; Scherer, 2003; Schroeder, 2001), although the evidence in this respect is inconclusive. Sadness and boredom, which reflect passive and resigned emotional states, have been accompanied by slower rates of speaking (Johnstone & Scherer, 2000; Scherer, 2003; Schroeder, 2001). Finally, a majority of studies investigating the effect of anxiety or worry on articulation rate reported an increase in speech rate when these emotions were associated with high emotional arousal (Siegman & Boyle, 1993; Siegman & Pope, 1965; Pope, Blass, Seigman, & Raher, 1970). However, the effects observed seem to depend on the subject’s susceptibility to and coping strategies (Johnstone & Scherer, 2000). Moreover, none of the studies were conducted with preschool-age children, an understudied population with respect to vocal expression of emotion. Nevertheless, articulation rate could be an emotion-specific acoustic parameter associated with speech fluency, a disruption of which characterizes stuttering; and to our knowledge, articulation rate has not been used as a potential marker of the intersection between speech-motor processes and emotion in young CWS.

1.4 Emotionality and Stuttering

Research suggests that temperamental characteristics and emotional processes are linked to stuttering in young children (a time marked by a rapid expansion of speech-language skills) (see the reviews by Conture et al., 2013; Jones, Choi et al., 2014; Kefalianos et al., 2012; cf. Alm, 2014). Although it is unclear whether emotional processes play a causal or consequential role in early stuttering, a number of studies have reported an association between the two (e.g. Ambrose et al., 2014; Arnold, Conture, Key, & Walden, 2011; Choi, Conture, Walden, Lambert, & Tumanova, 2013; Embrechts, Ebben, Franke, & Van de Poel, 2000; Walden, Frankel, Buhr, Johnson, Conture, & Karrass, 2012; Jones, Buhr, Conture, Tumanova, Walden, & Porges, 2014; Jones, Conture, Frankel, & Walden, 2014; cf. Reilly et al., 2009; Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014). These studies have used a variety of methods such as caregiver reports, behavioral observations (e.g., positive/negative expressions, attention shifts and fidgeting), and psychophysiology (e.g., heart rate or skin conductance).

Results indicate that young CWS exhibit lower adaptability to novelty and environmental change than non-stuttering children (Anderson, Pellowski, Conture, & Kelly, 2003; Embrechts et al., 2003; Howell, Davis, Patel, Cuniffe, Downing-Wilso, Au-Yeung, et al., 2004; Wakaba, 1998), increased emotional reactivity—that is, more frequent and higher levels of emotional arousal to emotional situations (Jones, Buhr et al., 2014; Karrass, Walden, Conture, Graham, Arnold, Hartfield, & Schwenk, 2006; Zengin-Bolatkale, Conture, & Walden, 2015)—and poorer emotion regulation—more difficulty regulating the occurrence, intensity or duration of emotional states (Jones, Buhr et al., 2014; Karrass, Walden, Conture, Graham, Arnold, Hartfield, & Schwenk, 2006). Others have reported that CWS, compared to CWNS, exhibit poorer attention regulation and inhibitory control (two important aspects of emotion regulation)—that is, less efficiency orienting, shifting or maintaining attention when appropriate, and less capacity to suppress inappropriate responses in novel or uncertain situations (Eggers, De Nil, & van den Bergh, 2010, 2012; Eggers, Luc, & van den Bergh, 2013; Felsenfeld, van Beijsterveldt., & Boomsma, 2010; Karrass et al., 2006; Schwenk, Conture, & Walden, 2007). Moreover, parent reports indicate that CWS have more negative mood (Eggers et al., 2010; Howell et al., 2004; Wakaba, 1998) and are more sensitive, anxious, fearful, shy, or withdrawn (e.g., Blood, & Blood, 2007; Fowlie & Cooper, 1978; Iverach et al., 2016). Behavioral reports indicate more negative affect in CWS, particularly in negative or frustrating situations (Johnson, Walden, Conture, & Karass, 2010; Ntourou, Conture, & Walden, 2013).

Empirical investigations exploring the link between stuttering and emotions have reported that preschool-age CWS exhibit more stuttering following emotionally arousing conditions (Johnson et al., 2010; Walden et al., 2012), especially when higher emotional arousal/reactivity is coupled with lower emotion regulation (Arnold et al., 2011; Walden et al., 2012; Jones, Conture et al. 2014; Jones, Walden, Conture, Erdemir, Lambert, & Porges, 2017). Only one longitudinal study has compared temperamental characteristics of preschool-age children who eventually recovered or persisted and found that children who persisted in stuttering were judged to have more negative affect by their parents than either those who recovered or their fluent peers (Ambrose et al., 2015).

1.5 Specific Purpose and Hypotheses

We know little about factors contributing to persistence and recovery from stuttering in young children; and no study, to our knowledge, has investigated whether articulation rate in relation to emotional processes differentiates children who are at higher risk of developing a chronic disorder from those likely to recover. We addressed three hypotheses relevant to this question.

First, we hypothesized that articulation rate of persisting, recovered and nonstuttering children would differ in response to three emotional conditions (positive, negative and baseline), as measured in *all types of utterances*—those that included stuttering-like disfluencies (SLD or SLD+OD utterance), non-stuttering-like (other) disfluencies (OD utterance), or were fluent (FLU utterance, containing no perceptible/discernable disfluencies).

Second, we hypothesized that articulation rate of persisting, recovered, and nonstuttering children would differ in response to the three emotional conditions, when articulation rate was measured in fluent utterances only (FLU utterance) with no perceptible/discernable disfluencies. Such a result would validate that the effect of emotion is present in fluent utterances that contained no carry-over effects from surrounding instances of stuttering.

Third, we hypothesized that articulation rate of persisting and recovered children would differ in response to the three emotional conditions when speech was *stuttered* versus *fluent*; since it is possible that the temporal coordination of speech segments in disfluent and fluent speech acts differently under the influence of emotion.

2. Methods

2.1 Participants

Participants were between 3;0 and 4;9 (months;years), and included 30 preschool-age children, who were classified as persisting (n=10), recovered (n=10), or nonstuttering (n=10) approximately 2–2.5 years after the initial diagnostic evaluation and experimental testing, when data for the current study was gathered. There were 9 boys and 1 girl in each group, matched in terms of age, with no significant differences in chronological age for persisting (M = 46.91 months, SD = 4.52), recovered (M = 46.11 months, SD = 6.87), or nonstuttering groups (M = 45.94 months, SD = 4.45). All participants demonstrated normal hearing on a bilateral tone hearing screening at pure tone frequencies of 1000, 2000, and 4000 Hz at 25 dB HL and were monolingual English speakers with parental reports of no history of neurological. Participants were referred to the study by their caregivers who were informed via a parent-oriented magazine, local health providers, or self/professional referral to the Vanderbilt Bill Wilkerson Hearing and Speech Center. Informed consent by parents and assent by children were obtained from all participants as part of the protocol for Vanderbilt University's Institutional Review Board. Participants were part of a larger group of over 200 participants in a longitudinal investigation of emotional and linguistic contributions to childhood stuttering conducted by Vanderbilt University's Developmental Stuttering Project (e.g., Choi et al., 2013; Clark, Conture, Walden, & Lambert, 2015; Jones, Buhr, et al., 2014; Ntourou et al., 2013).

The sample size for recovered and nonstuttering groups was determined based on the number of persistent participants from the initial data pool (n=10). Participants for the recovered and nonstuttering groups were selected randomly within the constraints of matching for age, gender and language scores. If a particular subject data skewed the balance of age, gender or language scores across the three groups, he/she was replaced by another randomly chosen subject.

2.2 Classification and Inclusion Criteria

Prior to experimental testing, children participated in a diagnostic evaluation in which they were evaluated by a speech-language pathologist, as they engaged in free play in which they provided a 300-word conversational speech sample. They were then administered several standardized speech-language assessments; and children below the 17th percentile on any test were eliminated to control for speech-language disorders. During this testing, participants were classified as CWS if they exhibited 3 or more stuttering-like disfluencies (SLDs; sound-syllable repetitions, monosyllabic whole-word repetitions, audible and inaudible sound prolongations) per 100 words of conversational speech (Conture, 2001; Yaruss, 1997) and scored 11 or higher (i.e., a severity equivalent of at least "mild") on the Stuttering Severity Instrument (SSI-3, Riley, 1994). Children were classified as CWNS if they exhibited 2 or fewer stuttered disfluencies per 100 words of conversational speech, and scored 10 or below on the SSI-3.

Persistency and recovery status was based on speech-language assessments at the participants' final diagnostic evaluation, approximately 2–2.5 years after the initial diagnostic evaluation and experimental testing when data for the current study was gathered. Each participant visited the lab 4–5 times (time points), each approximately 8 months apart over a 2–2.5 year period. Both diagnostic and experimental testing occurred at each time point. Participants who were classified as CWS at the *first* and the *last* diagnostic evaluation were included in the persisting group. Participants who were classified as CWS at the *first* diagnostic evaluation but classified as nonstuttering at the *second, third, fourth* or *last* diagnostic evaluation were included in the recovered group. Children in the recovered group recovered at different time points. Once recovery from stuttering was identified, it was verified by further visits, unless the participant recovered at his/her last time point. Similarly, the status of nonstuttering for the nonstuttering group was verified in subsequent visits to ensure that a child who did not meet criteria for stuttering in the first visit did not start stuttering later.

To assure an unbiased representation of group differences without potential linguistic confounds, the children in the three groups did not differ on scores on standardized speech and language assessments of articulation, receptive and expressive vocabulary, receptive and expressive language at the diagnostic evaluation. The tests included the Goldman Fristoe Test of Articulation (GFTA; Goldman & Fristoe, 2000), Peabody Picture Vocabulary Test (PPVT-III; Dunn & Dunn, 1997), the Expressive Vocabulary Test (EVT-2; Williams, 1997), and the Test of Early Language Development (TELD-3; Hresko, Reid, & Hammill, 1999). Planned comparisons using independent sample t-tests compared persisting and recovered groups, as well as CWS and CWNS, for each speech-language assessment. The persisting group did not differ from recovered group on standardized scores of GFTA (p= .42), PPVT (p= .12), EVT (p= .39), TELD-receptive (p= .10), TELD-expressive, (p= .95), or TELD-spoken (p= .48). CWS did not differ from CWNS on GFTA (p= .17), PPVT (p= .93), EVT (p= .22), TELD-receptive (p= .63), TELD-expressive, (p= .42), or TELD-spoken (p= .83).

To assure an unbiased representation of the two stuttering sub-groups without potential confounds of stuttering severity, persisting and recovered groups did not significantly differ in frequency of SLDs (p= .52) or SSI scores (SSI-3, Riley, 1994) at their first diagnostic visit (p= .88). The persisting group had an average of 9.93 SLDs per 100 words (SD=6.53) and an average SSI score of 19.3 (SD= 6.84). The recovered group had an average of 8.63 SLDs per 100 words (SD=4.01) and an average SSI score of 19.0 (SD=4.30). Individual participants' age and gender, frequency of SLDs and SSI scores from the initial screening, and time since onset (TSO) of stuttering reported by the parent at the initial time point appear in Table 1.

2.3 Procedure

Upon arrival at the Vanderbilt Developmental Stuttering Laboratory for experimental testing at the first time point, participants were seated in a car safety seat directly in front of a computer monitor. They watched three types of 4-min video clips (baseline, positive, and negative), and *immediately* after each clip they engaged in a narrative task. In the baseline condition, participants viewed a 4-min animated screensaver of a fish tank. Emotionally arousing film clips were selections of positive and negatively arousing scenes from one of five age-appropriate videos for children, including Happy Feet, The Lion King, The Little Mermaid, The Wizard of Oz, and The Princess and the Frog. The video clips were intended to elicit negative and positive emotional states. Trained research personnel validated that the video clips elicited the expected emotions by observing facial expressions while children watched the videotapes. Two research assistants blindly viewed videotapes of 8 CWS and 8 CWNS for each of the three experimental conditions and correctly identified the valence of the video clip 79% of the time.

Each child watched the baseline video clip first, and order of negative and positive video clip presentations was counterbalanced across children in each group to control for potential order effects. Following each clip, participants engaged in a narrative task, in which they told a story about one of three text-less storybooks about a boy, a dog, and a frog by the author Mercer Mayer (e.g., Frog Where Are You?; Mayer, 1969). The narratives were recorded on a desktop computer using a lapel microphone placed on the shirt of the child. The narratives were of a variety of lengths since each child spoke a different amount. We used the first 4 min of each narrative for data coding to match the length of the narratives. Although still limited in terms of its control over linguistic variability, storytelling based on the same picture book was thought to provide some control for confounds such as different levels of lexical, syntactic and structural complexity found in produced utterances, when compared to use of spontaneous conversational speech samples, as has been done in past research on articulation rate (Chon et al., 2012; Hall et al., 1999; Kloth et al., 1999; Turmanova et al., 2011).

2.4 Measures

2.4.1 Transcriptions of the utterances—A trained speech-language pathologist (SLP) orthographically transcribed utterances from the first 4 min of each narrative task using the

Systematic Analysis of Language Transcripts software program (SALT; Miller & Chapman, 2000). The transcriptions involved marking disfluencies, their types (e.g., sound-syllable repetitions), and the total number of syllables from fluent portions of utterances. SLDs are stuttered disfluencies, and following Tumanova et al. (2011), they included sound-syllable repetitions (SSR; e.g. “a-a-and”, “do-doing”), monosyllabic whole-word repetitions (WWR; e.g. “and-and”, “I-I-I”) and audible or inaudible sound prolongations (ASP or ISP; e.g. “aaaaa-and”, “ww-wanted”, “but—terfly”). Other disfluencies (OD) are non-stuttered (normal) disfluencies in the speech of fluent speakers, and following Tumanova et al. (2011), they included phrase repetitions (PR; e.g. “the frog – the frog jumps”), interjections (INT, e.g. “uhm”), and revisions (REV; e.g. “he is– they are leaving”). During the transcription each utterance was also categorized as one of four types following Chon et al. (2012): (1) FLU: perceptually fluent utterance that had no perceptible/discernible disfluencies, (2) SLD: stuttered utterance containing at least one SLD but no OD, (3) SLD +OD: stuttered utterance containing at least one SLD as well as at least one OD, and (4) OD: utterance containing at least one OD but no SLD. Total number of utterances transcribed and analyzed ranged from 10 to 39 in a given condition per participant. For inclusion in the analyses, the participants needed to contribute at least three stuttered (SLD and SLD+OD), or fluent (FLU) utterances in each condition.

The first author listened to the speech samples in their entirety while coding the utterances in Praat (Boersma, 2002), and rechecked the marking of disfluencies and the utterance categorizations, through ear and visual inspection of the spectrogram. If discrepancies appeared, they were resolved though repeated listening by the first author and the SLP, and reaching an agreement on categorization. If a discrepancy still remained, a second SLP listened to the sample for a final agreement. If no agreement converged the utterance was removed from the analyses.

For the first hypothesis on the effect of emotion on overall articulation rate, all four types of utterances were included in the analyses. For the second hypothesis on the effect of emotion on articulation rate in fluent speech, only the FLU utterances were included. For the third hypothesis on the effect of emotion on articulation rate of stuttered versus fluent utterances, the SLD and SLD+OD utterances for assessing stuttered speech, and the FLU utterances for assessing fluent speech were included.

2.4.2 Articulation rate—Articulation rate was defined as the number of syllables per second of perceptually fluent speech, removing all instances of stuttering and non-stuttering-like disfluencies and pauses greater than 250 ms (Chon et al., 2012, Hall et al., 1999; Walker, Archibald, Cherniak, & Fish, 1992; Yaruss, 1997). Utterances with fewer than three consecutive words, generic utterances not related to the content of the story (such as “I don’t know” or “turn the page”), as well as simultalk (where the speech segment of the child overlaps with that of the experimenter), unintelligible talk, and talk with a character voice (i.e., pretending to talk like a character) were excluded from analysis (Hall et al., 1999; Logan & Conture, 1995; Sawyer et al., 2008; Yaruss, 1997; Yaruss & Conture, 1995).

Praat speech analysis software (Boersma, 2002) was used to mark the onset and offset of each utterance, as well as the onset and offset of disfluencies and pauses longer than 250 ms within each utterance. Next, the duration of SLDs, ODs and pauses (if present) were subtracted from the overall duration of the utterance, yielding duration of perceptually fluent speech. This final duration was used to measure articulation rate in syllables per second. Accurate measurement of exact onset and offset points primarily depended on information from the waveform along with its corresponding spectrogram and amplitude envelope. The onset points were determined as the beginning of acoustic energy and offset points were determined as the termination of acoustic energy. Following Throneburg and Yairi (2001) and Chon et al. (2012), when measuring the duration of repetitions (sound-syllable repetitions [SSR], monosyllabic whole-word repetitions [WWR], and phrase repetitions [PR]), the entire portion of disfluency including all repetition units and pauses were included. When measuring audible sound prolongations [ASP], interjections (I), and revisions [REV] the whole duration of the disfluent period was measured. If prolongations happened as part of a multi-syllable word, only the affected syllable was marked as disfluent. For inaudible sound prolongations [ISP], the marked selection started with the point of first unnatural forced hesitation and ended with the end of disfluent speech. When two or more disfluency types happened in succession, representing disfluency clusters, the entire disfluent period including all pauses within the period were subtracted from the total duration. When silent periods were 250 ms or shorter they were retained, as they were considered a natural part of speech; those longer than 250 ms were subtracted (Andrews, Howie, Dozsa, & Guitar, 1982; Chon et al., 2012; Hall et al., 1999). Articulation rate was calculated for each utterance that conformed to the criterion for inclusion using the following formula:

$$\frac{\text{total duration of the utterance} - (\text{duration of disfluencies} + \text{duration of pauses} > 250ms)}{\#\text{ of syllables of the fluent portion}}$$

A Praat script was created to extract the relevant duration for measuring articulation rate as described in the above formula. Apart from the articulation rate, utterance length in syllables was also noted for each utterance to be used in the statistical analysis. Figure 1 provides a visual example of the analysis. The utterance “tttttttt-the frog jumps a-a- and tries to (get) grab the cup.” has one prolongation (“ttttttt-the”), one sound syllable repetition (“a- a- and”), and one revision (“(get) grab”). The utterance also contains two long pauses of 380 ms and 675 ms between words, which were excluded from the analysis. The durations marked in color were excluded from the total duration of the utterance, and speech rate was calculated on the remaining intact portion, which had eight syllables.

2.4.3 Measurement reliability—To assess measurement reliability for articulation rate, 20% of the total final data from each group was randomly selected for re-analysis to determine inter-rater reliability between the first author and a trained researcher (of doctorate level). Intra-class correlation coefficients (ICC; McGraw & Wong, 1996; Shrout & Fleiss, 1979) were computed for each group separately using the absolute agreement criterion. Correlations between the two raters were .977 (CI=.96–.99 $p < .0001$); .982 (CI=.97–.99 $p < .0001$); and .970 (CI=.93–.98 $p < .0001$) for persisting, recovered and nonstuttering groups, respectively.

2.5 Statistical Analysis

Three linear mixed-effects models (LMMs, Diggle, Heagerty, Liang, & Zeger, 2013; Pinheiro & Bates, 2000) addressed the three research questions using the mixed procedure of SPSS, version 25 (IBM Corp, Armonk, NY, USA). The LMM, compared to a standard ANOVA approach, is known to have better control for correlated residuals gathered from repeated measurements nested within and between individuals (Nich & Carroll, 1997). The models addressing the *first* and *second* hypotheses examined the effect of emotional condition on articulation rate (1) when articulation rate was calculated on all types of utterances and (2) when articulation rate was calculated on fluent utterances only. The two models included a between-subjects fixed factor of group (persisting, recovered, nonstuttering), a within-subjects fixed factor of emotional condition (baseline, positive, negative), and a within-subject fixed factor of utterance number (each child contributed a different number of utterances in the first 4 minutes), with utterance length (in number of syllables) as a covariate. The model addressing the *third* hypothesis examined the effect of emotions on articulation rate for fluent versus stuttered utterances. It included a between-subjects fixed factor of group (persisting, recovered), a within-subjects fixed factor of emotional condition (baseline, positive, negative), a within-subject fixed factor of utterance type (stuttering, fluent); and a within-subject fixed factor of utterance number, with utterance length (in number of syllables) as a covariate. Utterance length was covaried in each model to account for its possible effects on articulation rate, since it has been reported that longer utterances are produced at faster rates than shorter utterances (Malecott, Johnston, & Kizziar, 1972; Chon et al., 2012). This is because longer utterances place more demand on speech-motor control due to more complex linguistic and grammatical programming. Including utterance length as a covariate also accounts for findings that utterances with SLDs have been reported to be significantly longer than those that are perceptually fluent (Chon et al., 2012; Logan & Conture, 1995). In the linear models used, the intercept term was treated as a random effect that varies by individual to account for within-subject correlation of responses collected on a particular individual over time. Additionally, we used a time-based covariance structure to account for the spatial correlation among repeated utterances produced by a particular participant, as we predicted that the observations closer in time would be more similar than observations further apart.

3. Results

3.1 Analyses Regarding a Priori Hypotheses

Table 2 provides descriptive statistics for overall articulation rate, and articulation rate during fluent and stuttered utterances for each group.

3.1.1 Articulation rate during overall speech in three emotion conditions for persisting, recovered and nonstuttering children (Hypothesis 1)—The analysis of the first hypothesis showed a group by condition interaction, $F(4, 490.66) = 3.88$, $p = .004$; and a main effect of group, $F(2, 26.88) = 3.21$, $p = .056$, that approaches significance, when articulation rate was measured on all types of utterances. Figure 2 displays overall articulation rates for each group in each emotional condition. Overall, the persisting group was slower than the recovered group ($\beta = -.33$, $SE = .13$, $p = .018$), whereas the nonstuttering group did not differ from either persisting (p = .29) or recovered groups (p = .16).

Following up the interaction between group and condition, the persisting group was slower than the recovered ($\beta = -.52$, SE = .14, p = .002, Bonferroni adjusted) and nearly slower than the nonstuttering group ($\beta = -.34$, SE = .14, p = .06, Bonferroni adjusted) following the negative video clip. Similarly, the persisting group had significantly lower articulation rates following the negative clip compared with the baseline clip (β = .20, SE = .07, p = .009, Bonferroni adjusted), and compared with the positive clip (β = .26, SE = .07, p = .001, Bonferroni adjusted); unlike either recovered or nonstuttering groups whose articulation rates were not affected by the negative emotional condition (recovered, baseline vs. negative, p = 1; nonstuttering, baseline vs. negative, p = .766). In other words, persisting children slowed down in response to negative emotion, unlike recovered and nonstuttering groups that did not display any effect of emotion on articulation rate.

This analysis indicated that the borderline main effect of group was possibly driven by slower rates of the persisting group following negative emotion, as follow-up comparisons showed that during the baseline condition persisting, recovered and nonstuttering groups did not differ (persisting vs. recovered, p =.25; persisting vs. nonstuttering, p = 1; recovered vs. nonstuttering, p = .51, Bonferroni adjusted). Although the mean articulation rate of the recovered group was .34 syllables per second higher than that of the persisting group following the baseline condition, they did not differ when corrected for multiple comparisons.

3.1.2 Articulation rate during fluent speech in three emotion conditions for persisting, recovered and nonstuttering children (Hypothesis 2)—The analysis of the second hypothesis was parallel to that for the first hypothesis; with significant group differences, $F(2, 27.02) = 5.26$, p = .012; and of particular interest, a significant group by condition interaction, $F(4, 367.50) = 3.12$, p = .016, during fluent utterances. Overall the recovered group was faster than the persisting (β = .38, SE = .13, p = .007); and the nonstuttering groups (β = .34, SE = .13, p = .012). Consistent with the findings for the first hypothesis, during fluent speech following the negative video clip, the persisting group was slower than the recovered group ($\beta = -.63$, SE = .16, $p < .0001$, Bonferroni corrected), whereas the persisting and nonstuttering groups did not differ ($\beta = -.29$, SE = .15, p = .17, Bonferroni corrected). Similar to the findings on all types of utterances, the persisting group had significantly slower articulation rates during their fluent utterances following the negative emotion condition compared to baseline ($\beta = -.28$, SE = .10,, p = .028; Bonferroni corrected) and compared to positive ($\beta = -.32$, SE = .11,, p = .009; Bonferroni corrected), unlike either recovered or nonstuttering groups, whose articulation rates were not affected by the negative emotional conditions (recovered, baseline vs. negative, p = .782; nonstuttering, baseline vs. negative, p = .644). Figure 3 displays articulation rates during fluent speech for each of the three groups in each emotion condition.

3.1.3 Articulation rate during fluent vs. stuttered speech in three emotion conditions for persisting and recovered children (Hypothesis 3)—Finally, the third hypothesis addressed differences in articulation rate during fluent versus stuttered speech between persisting and recovered groups in three emotion conditions. The results showed a main effect of group, $F(1, 17.92) = 5.19$, $p = .035$, a group by condition interaction, $F(2, 297.92) = 3.13$, $p = .01$, and a group by utterance type interaction, $F(1, 917.34) = 7.84$, $p = .005$), with no significant three-way interaction ($p = .581$).

The group by condition interaction reflected the persisting group exhibiting slower articulation rate during speech following negative emotion as obtained for results of hypotheses 1 and 2. Therefore, we focused on the group by utterance type interaction. Follow-up analyses showed that both persisting ($\beta = .17$, $SE = .06$, $p = .008$; Bonferroni corrected) and recovered groups ($\beta = .43$, $SE = .066$, $p < .0001$; Bonferroni corrected) spoke at a faster rate during fluent than stuttered speech, but this difference was significantly more pronounced for the recovered than persisting groups ($\beta = .25$, $SE = .08$, $p = .005$). Additionally, there was no difference in articulation rates during stuttered speech between the two groups ($p = .244$); but the recovered group was faster than the persisting during fluent speech ($\beta = .42$, $SE = .14$, $p = .005$). Results from this analysis indicated that the main effect of group (also observed in the first two analyses) was primarily driven by fluent utterances of recovered children; and that when compared to persisting, the recovered group showed a more pronounced increase in articulation rate for fluent than stuttered speech. However, absence of a three-way interaction indicated that, contrary to expectations, the interaction was not moderated by different emotional conditions. Figure 4 shows articulation rates for stuttered and fluent utterances for persisting and recovered groups collapsed across emotional conditions.

4. Discussion

This study investigated the association of emotional processes and articulation rate in preschool age CWS who will eventually recover, CWS who will persist, and those who do not stutter. The study's main findings indicated that: (1) persisting children exhibited significantly slower articulation rates following a negative emotion induction, unlike recovered and nonstuttering children whose articulation rates were not affected by either of the two emotion-inducing conditions, (2) the same pattern was observed during fluent utterances that contained no carry-over effects from surrounding instances of stuttering, and (3) recovered children displayed significantly faster speech during fluent speech than during stuttered speech, when compared to the persisting group (although no effect of emotion was observed in this regard); suggesting a more pronounced disparity between speech-motor control of articulation rate for fluent versus stuttered speech for recovered children.

4.1 Effect of negative emotion on articulation rate in persisting stuttering

Only persisting children had a significant reduction in articulation rates following the negative-emotion eliciting video clip (Hypothesis 1). The 4-min negative video clips were intended to elicit negative emotional states, and the scenes were carefully chosen to portray fearful and unsettling situations from age-appropriate videos. It is important to note that at the time the speech samples were collected, there was virtually no difference between the persisting and recovered children in terms of stuttering severity (SSI score) and frequency of SLDs, or any of the speech language tests, including PPVT, EVT, TELD, and GFTA. Since the data used for this study was part of a longitudinal study, approximately 2–2.5 years after the initial testing these children were classified as either persisting or recovered. However, only persisting children (but not recovered or nonstuttering children) displayed a significant reduction in articulation rates after experiencing negative emotions. This might indicate a possible breakdown in speech motor performance following negative emotions for the persisting children.

Such reduction was also prominent during persisting children's fluent utterances, which contained no stuttering or non-stuttering like disfluencies; thereby had no carry-over effects from the surrounding instances of stuttering (Hypothesis 2). It is important to underscore that the effect observed in all types of utterances was also observed in fluent utterances only, since children with a relatively higher frequency of stuttering produce overall slower articulation rates, as there could be anticipatory, carry-over or overflow effects (Zebrowski, 1994) in the fluent segments preceding or following the stuttered segments in the utterances (Tumanova et al., 2011).

To our knowledge, this study is the first to look at the possible interaction of emotional processes and talker group effects (persisting CWS, recovered CWS, and CWNS) on articulation rate. Furthermore, literature comparing persisting and recovered children on emotional measures is scarce. One study attempted to link temperament to persistence/recovery, and its findings are consistent with ours. Ambrose et al. (2015) found persisting children aged 2–4 and tested within twelve months of the reported onset of stuttering to be more negative in mood than those who recovered, as shown in fear and soothability subscales of Children's Behavior Questionnaire (Rothbart et al., 2001). If indeed persisting children are more fearful and less soothable in general, the emotional valence of the negative video clips might have been stronger for the persisting children than for recovered or nonstuttering children; which might then have affected their speech-motor processes to control articulation rate to a greater extent.

Adults who stutter have been reported to have slower motor responses than those who do not stutter (Adams, 1984; Lanssen & Wieneke, 1987), in line with our current findings. However, based on studies investigating the effect of emotions on vocal parameters of speech, almost all of which involved adult participants, experiencing negative emotions such as fear results in faster speech articulatory movements (Ellgring & Scherer, 1996; Johnstone & Scherer, 2000; Scherer 2003; Schroeder, 2001). This is contrary to our findings for persisting children, as slower rates would be expected from states of relaxation, boredom or sadness but not from fear or anxiety. Thereby, negative emotions such as fear, seem to impact the speech-motor planning, programming or execution system to control articulation rate in a different way for persisting children.

There are several possible mechanisms that may explain how negative emotions differentially impact the articulation rate of persisting children:

1) *Differential physiological processes associated with negative emotion could be occurring for the persisting children during negative video clip (physiological mechanism).* One possibility is that persisting children, compared to recovered and non-stuttering children, could experience physiological responses during and following the negative viewing condition that in turn impacts their articulation rate.

Although there is no consensus on a unitary definition of “emotion,” physiological processes are commonly included in the definition by expert emotion researchers, in addition to the subjective feeling and perceptual-cognitive processes associated with it (e.g., Izard, 2010). While we acknowledge that physiological activity does not solely reflect emotion, and that articulation rate is not a direct measure of emotional processes; two studies have contributed to the hypothesis that physiological changes (in response to emotion inducing video clips) might impact articulation rates during the narratives. For example, Gilissen, Koolstra, Ijzendoom, Bakermans-Kraneenburg, and van der Veer (2007) used a similar paradigm, in which 3- and 4-year old children were shown fear-inducing scenes from child-appropriate film clips, while simultaneously their skin conductance (SCL) and heart rate variability (RMSSD) were recorded. They found that children responded to fear-inducing scenes with an increase in SCL—indexing higher sympathetic activity, emotional reactivity and arousal — and a decrease in RMSSD indexing lower parasympathetic activity. While there are no published longitudinal findings on physiological responses of children who persist to date, cross-sectional results from Jones, Buhr et al. (2014) using an identical paradigm to ours found that young CWS exhibit differential physiological responses during viewing and speaking conditions (e.g., co-activation or co-inhibition of sympathetic and parasympathetic activity during speech); which might indicate that CWS have less adaptive patterns of sympathetic and parasympathetic activity during emotionally challenging situations. Therefore, one likely mechanism that accounts for the impact of emotion on articulation rate is through changes in autonomic nervous system activity (for reviews see Johnstone & Scherer, 2000; Scherer, 1986; Scherer 2003). To this end, it is possible that differential autonomic nervous system responses of persisting children impact the functioning of the speech-motor system and result in decreases in articulation rate. In this case, the slower rate of speech may reflect a lag in the planning and/or execution of motor commands used for articulatory movements (Peters, Hulstijn, & Startkweather, 1989), or in the coordination and/or execution of individual motor subsystems involved in speech (Alfonso, Watson, & Baer, 1987; Peters & Starkweather, 1990) in response to physiological changes caused by negative emotional states.

Another related possibility is that negative emotion experienced by persisting children is perceived as threat and manifests itself as a “freezing response,” which is characterized by an inhibition of movement, well documented in adults in response to anticipatory anxiety (Alm, 2004; Sagaspe, Schwartz, & Vuilleumier, 2011). Such freezing response would be expected to transfer to speech that is characterized by an inhibition of motor activity related to vocalization (Alm, 2004); more unstable speech movement execution and coordination (van Lieshout, David, Lipski, & Namasivayam, 2014), or lags in reaction time (Hennessey, Dourado, & Beilby, 2014; van Lieshout et al., 2014). For example, there is evidence that perceived threat induced by fearful faces stimulates a freezing response (Sagaspe, Schwartz, & Vuilleumier, 2011), whereby emotional processes combine with motor inhibition, leading to prolonged reaction times. Recent work by van Lieshout et al. (2014) and Hennessey et al. (2014) found evidence for a slower speech-motor reaction time in response to threat words from the Emotional Stroop task in PWS compared to matched fluent speakers (PWNS);

pointing to an attentional bias to threat words (similar to that found in anxiety), which in turn impacts speech-motor execution processes. In support of this view, it has also been argued that PWS are influenced by a disposition to being more anxious in general (Craig & Tran, 2014; Iverach, Menzies, O'Brian, Packman, & Onslow, 2011; Mahr & Torosian, 1999), along with some evidence of anticipatory anxiety in PWS during speech related tasks accompanied by a paradoxical parasympathetic suppression of heart rate (Weber & Smith, 1990; Caruso, Chodzko-Zajko, Bidinger, & Sommers, 1994); which then, could translate into speech-motor planning/execution difficulties (van Lieshout et al., 2014).

Therefore, given that persisting children were also rated as more fearful and less soothable by their mothers (Ambrose et al., 2015), it is possible that the slower rates observed in persisting children in our study (who will represent the PWS group at older ages) are part of a freezing response that is characterized by a lag in preparing and/or executing speech-motor processes, in response to the emotion related changes in ANS. However, the current study was not designed to address such a possibility, and the idea that physiological autonomic nervous system responses are related to speech-motor planning and production processes that control articulation rate in persisting children much await further empirical study, where changes in articulation rate are assessed simultaneously with emotion-related physiological changes.

2) *Negative emotions may impact attentional and/or cognitive processes necessary for the speech-motor system differently for persisting children (attentional and/or cognitive mechanism).* The slower rates of speech for persisting children might reflect a lag in the central motor planning, preparation and programming (Conture et al., 1993; Postma & Kolk, 1993) due to attentional and/or cognitive resources being drawn away from speech and toward an emotional situation. This possibility would seem to be supported by numerous findings suggesting that lower attention shifting (Eggers et al., 2010; Schwenk et al., 2007), poorer attention regulation (Felsenfeld, van Beijsterveldt, & Boomsma, 2010; Karrass et al., 2006), less efficient attentional orienting (Eggers et al., 2012; cf. Johnson, Conture, & Walden, 2012), lower executive functions (Jones, Walden et al., 2017; Kraft, Ambrose, & Chon, 2014), and anomalous brain networks that suport attention (Chang et al., 2017) are associated with childhood stuttering.

In support of such view, it has also been proposed that the preparation and execution of skilled speech motor actions in PWS is vulnerable to interference and disruptions from cognitive, linguistic, or emotional demands (Namasivayam & van Lieshout, 2011; Peters, Hulstijn, & van Lieshout, 2000; van Lieshout et al., 2004). In the light of recent longitudinal results (Ambrose et al., 2015), negative aspects of emotions could be placing greater demand on an already vulnerable speech motor system possessed by persisting children, thereby leaving them with diminished resources to plan for and produce speech quickly. This would suggest that persisting children could be more vulnerable to the effects of negative emotions on speech-motor processes, which in turn puts them at higher risk to persist. As slower speech rates help PWS to coordinate articulatory movements more effectively and achieve fluency (Adams, Lewis, & Besozzi, 1973; Janssen & Wieneke, 1987), it could be argued that slower speech rate was an adaptive response on part of the speech-motor system of persisting children to increased processing load induced by negative emotion.

In support of this possibility, we did not observe an increase in the SLDs for the persisting children during the negative condition despite the obvious reduction in articulation rate. Past research has indicated a negative correlation between articulation rate and frequency of SLDs (Zebrowski, 1994; Tumanova et al., 2011), however in the current study, the persisting children did not stutter more or less during speech following the negative video-clip compared to baseline ($p = .64$) or compared to the positive emotion condition ($p = .69$). Also, there were no differences between the recovered and persisting children in their SLD counts overall ($p = .82$) or in any of the narratives following the three emotional films (condition by group interaction; $p =.92$). Thus, persisting and recovered children had comparable counts of disfluencies in all narrative tasks, which further may suggest that this was an adaptive response to promote fluency with a speech-motor system that is already vulnerable to the effects of negative emotion.

The finding that no effect of the positive video clip was observed on articulation rate for any of the three groups is also in agreement with previous research that showed no difference between CWS and CWNS in positive affect in parent reports (Eggers et al., 2010), no difference in positive emotional expressions upon receiving a desirable gift (Johnson et al., 2010), and no difference in positive affect expressed while engaging in baseline or frustrating tasks (Ntourou et al., 2013). It could be that the speech-motor systems of persisting children are particularly sensitive to the influences from negative emotions such as fear, as suggested by Ambrose et al.’s (2015) findings.

In sum, results relating to the first two hypotheses indicated that persisting children slow down their speech (regardless of the presence or absence of stuttering) after experiencing negative emotion, which indicates more vulnerability to negative emotions.

4.2 Relation between articulation rate and speech fluency in stuttering children

Persisting and recovered children were compared in terms of articulation rates during fluent and stuttered speech as a function of emotion (Hypothesis 3). In light of previous empirical reports (Chon at al., 2012; Logan & Conture, 1995; Yaruss & Conture 1996) we had predicted that persisting and recovered children might differ in terms of their articulation rates depending on whether the utterance is spoken fluently or not, and that such a relation might have been moderated by emotional differences between the two groups of children.

Contrary to the prediction, emotions did not play a role in this; however, recovered and persisting children diverged in their articulation rates during fluent compared to stuttered speech. Both groups of children spoke faster during fluent than stuttered utterances, as expected based on past research indicating a negative correlation between articulation rate and frequency of SLDs (Zebrowski, 1994; Tumanova et al., 2011). Even though articulation rates were calculated on intact portions of utterances (removing disfluencies and pauses longer than 250 ms), the presence of disfluencies might still affect the articulation rates as there would be anticipatory and carry over effects (Zebrowski, 1994) in the fluent segments preceding or following the stuttered segments (Tumanova et al., 2011). In line with this, some researchers (Chon et al., 2012; Logan & Conture, 1995; Yaruss & Conture, 1996,) have reported slightly faster articulation rates for fluent than stuttered speech in CWS, however the differences were not statistically significant in those studies. Similarly the study by Chon et al. (2014) compared FLU, SLD, SLD+OD and OD types of utterances in pairs but reported no differences in the articulation rates between FLU and other types of utterances. On the contrary, this study compared FLU to SLD and SLD+OD combined, thereby comparing fluent utterances to stuttered utterances containing at least one SLD regardless of whether it contained any OD. The current results are consistent with previous empirical reports finding faster rates for fluent than stuttered utterances in CWS. This suggests that the speech-motor control system acts differently for the temporal coordination of speech segments in fluent and disfluent speech in CWS.

Of particular interest, in this study the difference in rate for fluent versus stuttered speech was more pronounced for recovered than persisting children, such that recovered children produced speech at a faster rate when it was fluent. This might indicate that recovered children use temporal aspects of speech motor control more efficiently than persisting children when producing fluent speech. This view is in line with the idea that faster speech develops as children mature (Logan, Bryd, Mazzocchi, & Gillam, 2011). Faster rates for adults reflect a growing maturity of the speech-motor system, as well as superior cognitive and linguistics abilities (Malecott, Johnson, & Kizziar, 1972). Similarly, faster rates of fluent speech for recovered children might reflect a more mature speech-motor system that is capable of producing motor commands, controlling timing of motor systems, or moving articulators more efficiently than persisting children.

It is also noteworthy that aside from the effect of negative emotion, stuttered utterances seemed to exert similar levels of demand for both persisting and recovered children as evidenced by their similar articulation rates during stuttered utterances. The difference was primarily driven by the production of fluent utterances, where it seemed more demanding for persisting children to plan for and produce fluent speech at a fast rate. Such demand might reflect greater efforts in executing motor commands, in controlling timing of individual motor systems, or in executing motor planning, thereby resulting in a lag in the speech motor output and slower articulation rates during fluent speech for the persisting children.

This is also in line with some converging lines of research, which argues for limited speech-motor skills in PWS compared to PWNS. PWS seem to benefit less from practice and seem to be less efficient and less flexible adapting to motoric and cognitive-linguistic task requirements, such as speaking rate changes (De Nil, 2004; van Lieshout et al., 2004). Similarly, it appears that the speech motor system of persisting children may be less able to maintain a fast pace while simultaneously coping with concurrent linguistic-cognitive demands. In this case, higher articulation rates in the case of recovered children could be viewed as a reflection of a more mature speech-motor system, which might play a role in promoting subsequent recovery from stuttering.

Present results suggest a more pronounced disparity between the mechanisms responsible for fluent and stuttered speech of recovered children. It would seem that recovered children possess a more flexible speech-motor system that adapts to faster rates more effectively under the absence of disfluencies, and a speech-motor system that is more vulnerable to co-occurring processes such as emotions for persisting children. More studies need to be conducted to validate the results and unravel the underlying mechanisms contributing to this disparity.

4.3 General considerations and implications

An important potential implication of our findings is that different underlying emotional and/or speech-motor processes could contribute to stuttering for persisting and recovered children. Stuttering is known to be a complex and multifaceted disorder, with linguistic/language factors, speech-motor factors, physiological factors, cognitive factors, and temperamental/emotional factors all playing potentially significant roles (e.g., Conture & Walden, 2012; Guitar, 2013; Smith & Kelly, 1997; Smith, 1999; Yairi & Ambrose, 2005). However, the present findings suggest that negative temperamental/emotional factors might play a more critical role in the speech-motor execution of speech for children who develop stuttering and persist than for those who recover. In the case of persistent stuttering, negative emotions might play a more complicated role by interacting with the speech-motor execution system, whereas for children who recover from stuttering, stuttering might reflect a temporary speech-motor problem in and of itself without vulnerability to disruption by emotion.

Notably, we also observed no change in articulation rate for the stuttering children who later recovered or nonstuttering children in response to emotional manipulations. Although past research has shown a difference between CWS and CWNS in terms of emotional reactivity and regulation (Eggers et al., 2010, 2012, 2013; Felsenfeld et al., 2010; Jones, Buhr et al., 2014; Karrass et al., 2006; Schwenk et al., 2007; Zengin-Bolatkale et al., 2015), as well as differences in negative aspects of emotionality (Eggers et al., 2010; Howell et al., 2004; Johnson et al., 2010; Ntourou et al., 2013; Wakaba 1998), none of these studies grouped CWS based on persistence and recovery, as it would require repeated testing at multiple time points spread out. Therefore, the differences observed in emotion and temperament between CWS and CWNS in past research might have been produced at least in part by the stuttering children who would persist. Such a view may indicate that the mechanisms that lead to stuttering in children who eventually recover and in children whose stuttering persists operate differently.

From a clinical perspective, our results also suggest that articulation rates in response to negative emotion, as well as articulation rates during stuttered versus fluent speech might be considered as potential factors to prospectively predict persistence and recovery from stuttering, and identify those at high risk to provide optimal treatment in a timely manner. Understanding the disparity between the recovered and persisting children in terms of their speech-motor maturity and flexibility to be able to handle higher articulation rates during fluent speech also has clinical relevance for speech language pathology.

5. Summary and Conclusions

In summary, findings from this study showed that only children whose stuttering would eventually persist slow down significantly when experiencing negative emotion, suggesting that articulation rate, when considered in relation to emotionality, might be a potential marker for identifying children who are at higher risk of chronic stuttering. Moreover, higher rates of fluent speech compared to stuttered speech for the children who would eventually recover may indicate a disparity in the speech-motor mechanism for controlling fluent versus stuttered speech, which may suggest that recovered children have a more mature speech-motor system that is capable of preparing for and performing motor actions more efficiently in fluent speech. Together the results seem to suggest salient differences in the underlying speech-motor processes that contribute to stuttering chronicity, such that emotion and stuttering differentially impact articulation rate of persisting compared to recovered children. Ultimately, these results highlight the importance of considering multiple factors to develop a comprehensive understanding of the developmental trajectory of childhood stuttering.

Acknowledgments

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), as well as the National Center for Research Resources, a CTSA grant (1 UL1 RR024975–01) to Vanderbilt University that is now at the National Center for Advancing Translational Sciences (UL1 TR000445–06). The research reported herein does not reflect the views of the NIH, NIDCD, or Vanderbilt University. The authors would like to thank the late Dr. Warren Lambert for help with statistical analyses. We also extend sincere appreciation to the young children and caregivers whose participation and cooperation made this project possible.

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Highlights

- Children who would persist in stuttering exhibit slower articulation rates following negative emotion unlike recovered or nonstuttering children.
- Stuttering children overall exhibit faster articulation rates during fluent compared to stuttered utterances.
- Compared to persisting, recovered children exhibit significantly faster rates in fluent speech at a time when there is no subtype classification.
- Differences in articulation rate in pre-school age children who stutter may be linked to differences in emotional processes.
- Articulation rate could be a potential factor to prospectively predict persistence and recovery from stuttering.

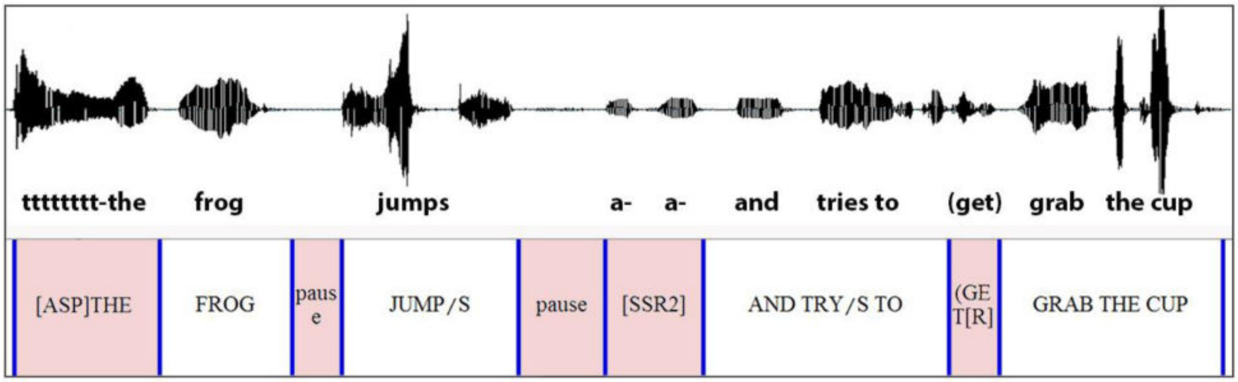

Figure 1.

Visual example of the analysis. Utterance, “ttttttt-the frog jumps a-a- and tries to (get) grab the cup” containing two SLDs (one audible sound prolongation, and one sound-syllable repetition), one OD (revision), two pauses longer than 250 ms, and 8 fluent syllables.

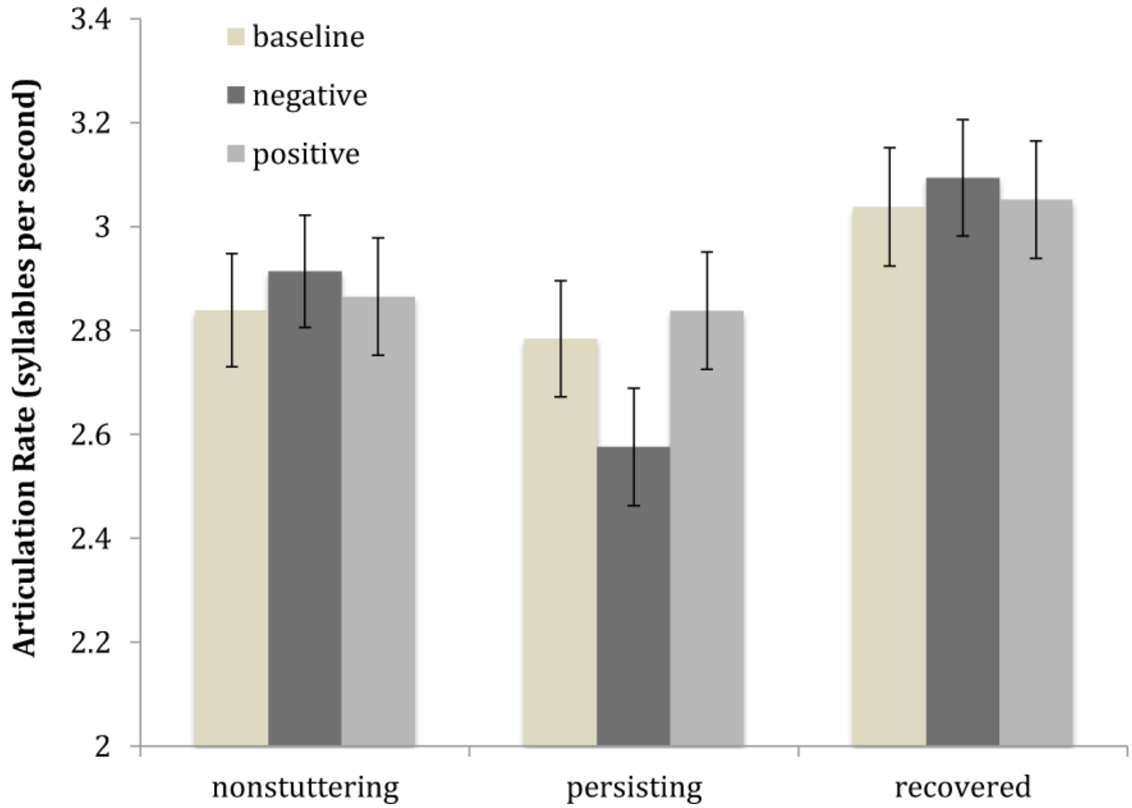

Figure 2.
Articulation rates as measured on all types of utterances, for each of the three groups, in each of the three emotional conditions, controlling for utterance length. Error bars represent standard error of the mean.

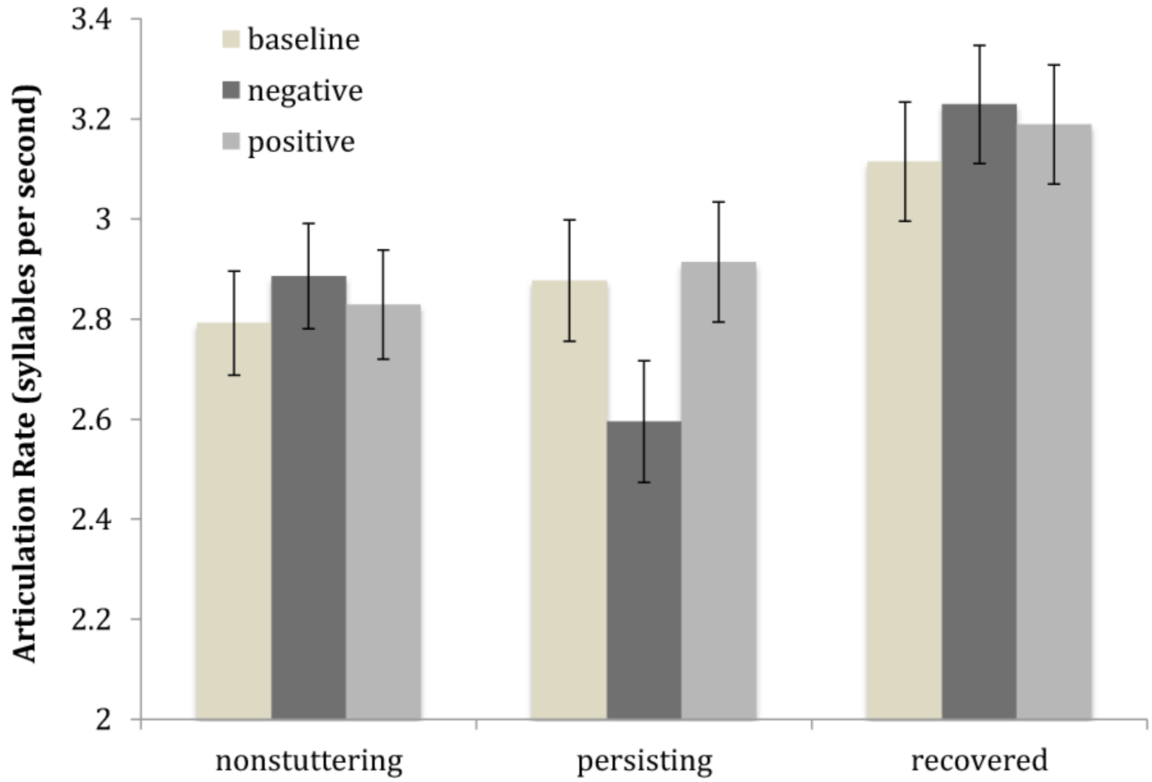

Figure 3.

Articulation rates measured during fluent utterances only for each group in each of three emotional conditions, controlling for utterance length. Error bars represent standard error of the mean.

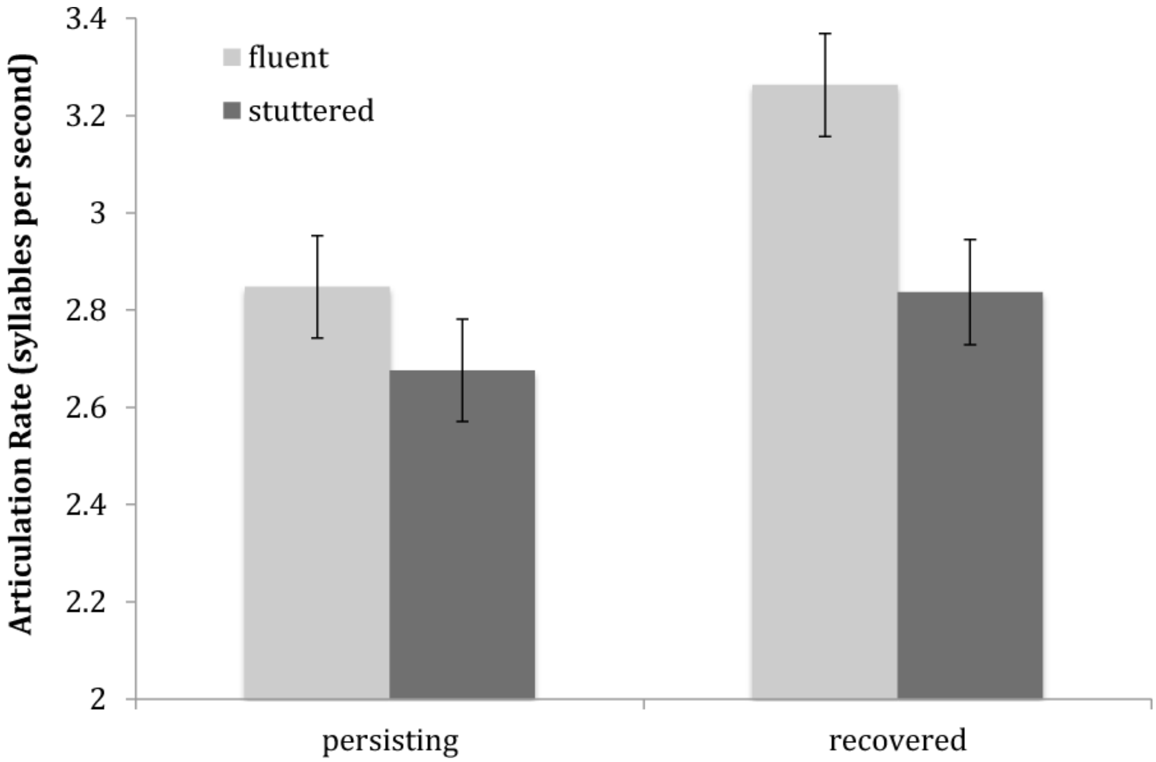

Figure 4.
Articulation rates during stuttered versus fluent utterances for persisting and recovered groups collapsed across the three emotional conditions. Error bars represent standard error of the mean.

Table 1

Age and gender for each participant in each group, frequency of SLDs and SSI scores from the initial screening, and TSO of stuttering as reported by the parent at the initial time point.

	Gender	Age in months	SLDs per 100 words	SSI score	TSO at their first time point (in months)
Persisting					
P01	M	48.8	22.04	27	9
P02	M	43.4	10.33	22	10
P03	M	50	5.33	14	20
P04	M	37.7	4.67	14	4
P05	M	45.9	6	16	4
P06	M	44.9	18.67	22	10
P07	M	46	15.33	33	9
P08	M	46.9	4.33	13	11
P09	F	52.9	4	12	16
P10	M	52.6	8.67	20	13
Mean (SD)	—	*46.9 (4.5)*	*9.9(6.5)*	*19.3(6.8)*	*10.6 (4.9)*
Recovered					
R01	M	43	13.67	24	11
R02	M	40.5	5	14	4
R03	M	56.6	7.67	20	9
R04	M	36.7	9.67	24	9
R05	M	50	10.33	18	11
R06	M	49.3	9.33	23	1
R07	M	45.6	4.67	16	3
R08	F	49.1	4	12	6
R09	M	54	3.67	14	12
R10	M	36.3	15.33	24	11
Mean (SD)	—	*46.1 (6.9)*	*8.3 (4.0)*	*18.9(4.7)*	*7.7 (3.9)*
Nonstuttering					
N01	M	39.2	2	8	—
N02	M	45.2	1	6	—

	Gender	Age in months	SLDs per 100 words	SSI score	TSO at their first time point (in months)
N03	M	46.9	1.33	6	—
N04	M	50.9	1.33	6	—
N05	M	48.3	0.33	6	—
N06	M	42	1.33	8	—
N07	F	46.8	2	10	—
N08	M	52.7	1	7	—
N09	M	48.3	1.33	6	—
N10	M	39.1	2	8	—
Mean (SD)	—	*45.9 (4.6)*	*1.4(0.5)*	*7.1 (1.3)*	—

Note: SD = standard deviation; M = male; F = female; SLD = stuttering like disfluencies; SSI score = stuttering severity instrument (Riley, 1994); TSO = Time since onset of stuttering.

Table 2

Means (M) and standard deviations (SD) for articulation rate (number of syllables per sec) for the persisting, recovered and nonstuttering groups.

Conditions	Articulation rate overall	Articulation rate during *fluent* utterances	Articulation rate during *stuttered* utterances
	M (SD)	***M (SD)***	***M (SD)***
Persisting			
Baseline	2.75 (0.69)	2.84 (0.72)	2.69 (0.67)
Negative	2.57 (0.53)	2.58 (0.49)	2.52 (0.53)
Positive	2.85 (0.72)	2.92 (0.79)	2.76 (0.66)
Recovered			
Baseline	3.05 (0.76)	3.17 (0.78)	2.91 (0.74)
Negative	3.09 (0.81)	3.28 (0.82)	2.83 (0.68)
Positive	3.05 (0.70)	3.22 (0.71)	2.83 (0.66)
Nonstuttering			
Baseline	2.83 (0.73)	2.84 (0.74)	
Negative	2.83 (0.65)	2.85 (0.64)	
Positive	2.80 (0.69)	2.85 (0.71)	

Note: For the purposes of description, this table presents raw means and standard deviations, uncorrected for the covariate (utterance length).