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Articulation rate and its relationship to disfluency type, duration, and temperament in preschool children who stutter

Victoria Tumanova,

Department of Communication Sciences and Disorders, The University of Iowa, Wendell Johnson Speech and Hearing Center, Iowa City, IA 52242, phone: +1 319 335-8738, fax: +1 319 335-8851

Patricia M. Zebrowski,

Department of Communication Sciences and Disorders, The University of Iowa, Wendell Johnson Speech and Hearing Center, Iowa City, IA 52242, phone: +1 319 335-8735

Rebecca N. Throneburg, and

Department of Communication Disorders and Sciences, Eastern Illinois University, Human Services Center, 600 Lincoln Ave, Charleston, IL 61920, phone: +1 217 581-2712

Mavis E. Kulak Kayikci

Hacettepe University, Faculty of Medicine, Ear, Nose and Throat Department, Audiology and Speech Pathology Unit, 06100 Sihhiye, Ankara, Turkey, phone: +90 312 305-4387,

Victoria Tumanova: victoria-tumanova@uiowa.edu; Patricia M. Zebrowski: tricia-zebrowski@uiowa.edu; Rebecca N. Throneburg: rmthroneburg@eiu.edu; Mavis E. Kulak Kayikci: mavis@hacettepe.edu.tr

Abstract

The purpose of this study was to examine the relationship between articulation rate, frequency and duration of disfluencies of different types, and temperament in preschool children who stutter (CWS). In spontaneous speech samples from 19 children CWS (mean age = 3:9; years: months), we measured articulation rate, the frequency and duration of (a) sound prolongations; (b) soundsyllable repetitions; (c) single syllable whole word repetitions; and (d) clusters. Temperament was assessed with the *Children's Behavior Questionnaire* (Rothbart et al., 2001). There was a significant negative correlation between articulation rate and average duration of sound prolongations (p<0.01), and between articulation rate and frequency of stuttering-like disfluencies $(SLDs)$ ($p<0.05$). No other relationships proved statistically significant. Results do not support models of stuttering development that implicate particular characteristics of temperament as proximal contributors to stuttering; however, this is likely due to the fact that current methods, including the ones used in the present study, do not allow for the identification of a functional relationship between temperament and speech production. Findings do indicate that for some CWS, relatively longer sound prolongations co-occur with relatively slower speech rate, which suggests that sound prolongations, across a range of durations, may represent a distinct type of SLD, not just in their obvious perceptual characteristics, but in their potential influence on overall speech production at multiple levels.

Correspondence to: Victoria Tumanova, victoria-tumanova@uiowa.edu.

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childhood stuttering; speech; articulation rate; disfluencies; prolongations; temperament

1. Introduction

Contemporary theories of developmental stuttering view the disorder as the product of a dynamic, nonlinear interaction of a number of risk factors (e.g. Conture et al., 2006; Ludlow & Loucks, 2003; Smith & Kelly, 1997). This concept of stuttering as a multifactorial disorder has been the impetus for moving both researchers and clinicians toward a theoretical framework that assumes that there is no single constitutional or environmental factor that is either necessary or sufficient for stuttering to emerge, or that underlies the patterns of persistence or recovery that have been observed in stuttering at its onset. Rather, it is the complex interaction of specific and perhaps idiosyncratic factors leading to a socalled "tipping point" that trigger the first observable behaviors that listeners consider to be stuttering (e.g. sound-syllable repetitions and sound prolongations; associated or secondary behaviors), as well as the non-observable cognitive and affective features that co-occur with these observable behaviors (e.g. speech and situational avoidance; social anxiety) (Davis, Shisca, & Howell, 2007). Such a multidimensional view aligns well with what is known about the nature of any complex human behavior (Thelen & Smith, 1994); at the same time, it obviously complicates attempts to understand stuttering from both research and clinical perspectives.

With this in mind, a fruitful avenue for research in early stuttering involves the identification of specific risk factors whose complex interactions compose subgroups within the general population of children who stutter. Earlier work by Preus (1981), Prins and Lohr (1972), and others suggested that the large degree of variability within the stuttering population in a number of dimensions is due to the existence of subgroups. In support of this view, Schwartz and Conture (1988), observed that subgroups of CWS could be identified by three main behavioral measures: the proportion of the total number of stuttered disruptions that are judged to be sound prolongations, or Sound Prolongation Index (SPI), the average number of associated or secondary behaviors the child produces, and the variety of different associated behaviors. More recently Yairi and his colleagues have published a number of studies which lend additional support for the existence of subgroups (for review see Yairi & Ambrose, 2005). Overall, research has led to the identification of a narrower set of risk factors that among others include the child's speech and language planning and speech production abilities, type of speech disfluency, and the child's temperament (for review, see Yairi, 2007).

In their "Communication-Emotional (CE)" model of stuttering, Conture, Walden, Arnold, Graham, Hartfield and Karrass (2006) have classified risk factors according to their distal or proximal contributions to stuttering and its development over time. The CE model considers the interaction between a child's genetic make-up and his or her daily environment as the key downstream, or distal, factor in the emergence of stuttering. That is, the way in which the child's inherited skills and abilities interact with the environment serves as the precursor, or foundation, for stuttering onset. Once stuttering has emerged, so-called upstream or proximal factors, typically subtle, observed at one or more levels of speech-language planning and production serve to trigger instances of stuttering (see Levelt, 1989 for model of different levels of speech production). Simply put, the interaction of genetics and environment create a "unique environment" for each child, one that can predispose the child to stutter; however, it is the proximal contribution of disruptions in speech and language planning and production that yield overt stuttering behavior. Finally, the development of a

persistent stuttering problem is predicated on the child's level of emotional reactivity and regulation. That is, the way in which a child reacts emotionally to his own stuttering behavior, and the extent to which he can regulate that reaction, will impact the frequency and perhaps the chronicity of stuttering.

That being said, a key question for researchers and clinicians alike is: What are relevant risk factors, and how do they relate to one another? As a beginning attempt to answer this question, we used both the Multifactorial (i.e. Smith and Kelly, 1997) and CE models as a framework for examining potential relationships between variables that both have identified as potential risk factors in the onset and development of stuttering. Both models, but in particular CE, suggest that the ways in which a child's stuttering characteristics interact with speech and language planning abilities and temperament may play a role in stuttering. Converging data from research in articulation rate, the type and duration of stuttered disfluencies, as well as temperament in young children who stutter supports the existence of such an interaction. The following sections provide a summary of research findings from studies of each of these variables, followed by a brief discussion of the how they may interact and the implications of this interaction for CWS.

1.1. Frequency and Duration of SLDs in CWS

Researchers and clinicians have long viewed sound prolongations as a sign of stuttering chronicity and severity, and a predictive factor in stuttering development. For example, Gregory (1973) described prolongations as atypical or "more unusual" disfluencies because he observed that they were relatively infrequent in the speech of normally disfluent children and more characteristic of what listeners perceive as "stuttering". This observation was corroborated by Yaruss, LaSalle, & Conture (1998) who analyzed the diagnostic data of a large number of CWS and concluded that the "sound prolongation index", or SPI (i.e. proportion of sound prolongations in all stuttering-like disfluencies) is a measure that contributes to perceptual judgment of stuttering severity (i.e., the higher SPI relates to judgments of more severe stuttering). It is noteworthy that clinician judgments of both the duration and frequency of sound prolongations are a key component in prognostic instruments designed to differentiate children at risk for continuing to stutter from those who are likely to recover on their own. Both the *Stuttering Prediction Instrument for Young Children* (Riley, 1981) and *The Chronicity Prediction Checklist* (Cooper & Cooper, 1985) list (among other criteria) the presence of prolongations and "blocks", as well as prolongations in excess of one second, as risk factors for the development of chronic stuttering.

Conture (1990), Curlee (1980) and Van Riper (1982) have all proposed that if early stuttering is dominated by sound prolongations as opposed to sound-syllable repetitions, the chances for unassisted recovery are smaller. Throneburg & Yairi (2001) examined this notion directly by measuring changes in both disfluency type and duration in two groups of CWS, one that developed chronic stuttering and the other that experienced unassisted recovery. They observed that children in the recovered group showed a change from a relatively high proportion of sound prolongations (aka "dysrhythmic phonations") to monosyllabic whole-word repetitions over time, whereas the children who persisted did not show such a pattern of change in disfluency type. Moreover, there was a significant change across visits in the proportional occurrence of disrhythmic phonations for the recovered group.

Along with the production of sound prolongations, duration of SLDs has long been thought to contribute to the identification and severity of stuttering in children. Presently, there are few published studies that support this conclusion. In fact, Zebrowski (1991) and Kelly and Conture (1992) observed that there was no significant difference between preschool CWS

and CWNS in the duration of either sound-syllable repetitions or sound prolongations. Zebrowski (1991) proposed that the duration of SLDs may not contribute to the differential diagnosis as much as proportion of disfluency type, but instead its significance may lie in the way it interacts with other dimensions of speech and stuttering.

1.2. Articulation Rate in CWS

Articulation rate in fluent speech has long been considered a measure of speech motor execution, in that it reflects the speaker's ability to temporally coordinate respiratory, phonatory and articulation processes (Hall, Amir, & Yairi, 1999; McClean, & Tasko, 2003; Tasko, McClean, & Runyan, 2007). From this perspective, slow articulation rate may signify either an immature or compromised speech motor control system. As such, researchers have speculated that CWS, as a group, produce slower articulation rates than their nonstuttering peers; however, findings from studies of rate have been equivocal. For example, while Meyers and Freeman (1985) observed that CWS spoke at a slower rate than children who do not stutter (CWNS), Kelly and Conture (1992) and Ryan (1992) reported no significant differences in articulation rate between these two groups (see Sawyer, 2008, for review). In addition to considering articulation rate as a measure of speech motor skill, psycholinguistic models of stuttering have implicated articulation rate as a contributing factor to fluency breakdowns in speech of people who stutter (Karniol, 1995; Perkins, Kent, & Curlee, 1991; Postma, & Kolk, 1993). These models have suggested that people who stutter require additional time for phonological processing and planning of speech movements, which hypothetically could result in slower articulation rate than that of people who do not stutter.

Several researchers have entertained the possibility that the significance of articulation rate is that it might be a predictor of either recovery or persistence in childhood stuttering. For example, Kloth, Janssen, Kraaimaat and Brutten (1995) measured articulation rate in normally fluent preschool children at risk to develop stuttering because of a positive family history. They found that the pre-onset articulation rate of 26 children who were considered to stutter at one year follow-up was significantly faster than that of 67 children who did not develop stuttering during that time period. In a later study, Hall, Amir & Yairi (1999) examined articulation rate in CWNS and two subgroups of CWS: those who developed persistent stuttering and those who recovered without intervention. They employed two metrics of articulation rate, namely, phones per second and syllables per second. Results showed that the recovered group spoke significantly slower than both the persistent CWS and the group of CWNS, when phones per second was the dependent variable. There were no significant differences between any of the groups when rate was measured in syllables per second (the conventional measure of articulation rate). The authors took this observation to indicate that phones per second was perhaps a more sensitive measure of speaking rate, and that children who recovered from stuttering without therapy were helped in this process by maintaining a slower rate of articulation. These disparate findings suggest that articulation rate, in and of itself, does not distinguish stuttering from nonstuttering children, its relevance, however, may lie in its relationship to stuttering development.

1.3. The Relationship between the Frequency and Duration of SLDs, and Articulation Rate in CWS

In 1994, Zebrowski published the results of a preliminary investigation that examined the extent to which articulation rate in fluent speech was related to specific characteristics of stuttered speech in a group of CWS. Measures of articulation rate, frequency and type of disfluency (including SPI), duration of sound prolongations and sound-syllable repetitions, and number and rate of repeated units per instance of sound and syllable repetition were obtained from samples of parent-child conversation. The results showed a significant positive correlation between sound prolongation duration and SPI, so that children who

prolongations, and a significant negative correlation between sound prolongation duration and articulation rate. That is, children who produced longer sound prolongations also produced slower articulation rate. This was not the case for articulation rate and the duration of sound-syllable repetitions. An additional finding was a significant negative correlation between the duration of sound-syllable repetitions and the rate, not the number, of repeated units, that is, longer durations of sound-syllable repetitions were associated with a slower rate of iteration and shorter durations with a faster rate of iteration, whereas the number of units being repeated in a sound-syllable repetition stayed fairly constant. This observation suggests that CWS change the duration of sound and syllable repetitions by manipulating the rate at which they produce the repeated units (faster or to increasing or decreasing the number of repeated units within the disfluent production.

Zebrowski interpreted these results as support for the numerous clinical reports that have argued for the significance of sound prolongation as an indication of progression and severity of stuttering in children. Specifically, she speculated that CWS who produce higher proportions of sound prolongations (i.e. larger SPIs) may exhibit inefficient strategies for making transitional gestures between speech movements during their fluent as well as stuttered speech. This in turn might lead to an "overflow" of the relatively fixed or arrested speech production strategy underlying prolonged sounds throughout the entirety of their speech, both fluent and disfluent. That is, increases in these relatively fixed transitional gestures (sound-to-sound) may lead to increased duration of sound prolongations and slower articulation rate. Similarly, this same strategy may be observed for sound-syllable repetitions as well, in that there is a negative relationship between rate of repeated units and duration (i.e. reduced rate of repeated units and longer sound-syllable repetitions).

Zebrowski, Conture (1990) and others have speculated that one of the bases for the emergence and development of prolonged sounds in the speech of CWS is the child's reaction to, and attempt to physically compensate for, earlier forms of stuttering (repetitions of sounds and syllables) by using fixed or physically tense speech production. In his "Alpha-Delta" hypothesis of the developmental progression of stuttering behaviors, Conture (1990) went so far as to suggest that reducing the rate of repeated units in sound-syllable repetitions may reflect a transitional stage between mainly repetitive (Beta) and primarily fixed (Gamma) stuttering behaviors; a phenomenon that may result in slowed articulation rate in fluent speech as well. A key question here is this: What is the origin and nature of the child's reaction to his own stuttering that would lead some CWS, and not others, to compensate for stuttering in this way?

One answer may lie in the CE Model of stuttering discussed earlier. The predictions of the theoretical model proposed by Conture et al. (2006), along with empirical evidence, suggests that some CWS possess a temperamental profile that may be a factor in the exacerbation of stuttering that manifests as changes in such features as duration and rate.

1.4. Temperament of CWS

Recently, several studies have examined the temperamental characteristics of young children and adolescents who stutter (Anderson, Pellowski, Conture, & Kelly, 2003; Davis, Shisca, & Howell, 2007; Embrechts, Ebben, Franke & van der Poel (2000); Karrass, Walden, Conture, Graham, Arnold, & Hartfield, 2006; Schwenk, Conture, & Walden, 2007). Anderson et al. and Karass et al. used a norm-reference parent questionnaire (McDevitt & Carey, 1978) to examine the temperament constructs of emotional reactivity and emotional regulation in young children who do and do not stutter (CWS and CWNS, respectively). Embrechts et al. (2000), also used parent report to assess the temperament in stuttering and nonstuttering children within this range of ages (3–7). Results of these studies indicated that children who

stutter were significantly more reactive and less able to regulate both emotionality and attention as compared to their normally fluent peers when controlling for gender, age, and language abilities. An additional, but related, finding from Embrechts et al. was that CWS were more likely than CWNS to exhibit higher levels of gross motor activity. In a later study, Schwenk et al. (2007) investigated the maintenance of attention and adaptation to background stimuli in three to five year-old CWS and CWNS. Results indicated that CWS were significantly more likely than CWNS to attend to, or look at changes in background stimuli, although there were no significant differences between the groups in duration or latency of this behavior. The findings were interpreted to suggest that preschool CWS are more reactive to, distracted by, and slower to adapt and habituate to environmental stimuli. Taken together, the findings from studies in the temperament of CWS have consistently shown that compared to CWNS, these children are more reactive in general and slower to self-regulate and adapt across behavioral domains.

1.5. The Relationship between Temperament and the Frequency and Duration of SLDs in CWS

Based on the results from studies of temperament and speech and language processing in CWS, Conture et al. (2006) have suggested specific relationships between these two factors. In their CE model of stuttering development, the authors propose that a complex interaction between speech and language processing, emotional reactivity, emotional regulation and attention regulation contributes in both distal and proximal ways to childhoods stuttering. For example, children between two and seven years of age are in the process of skill acquisition, including that necessary for speech-language planning and production. It has been well established that during normal early development, children exhibit relatively variable, perhaps unstable, planning and production within speech and language domains, and that variability is influenced by such factors as language complexity and age (e.g. Goffman and Smith, 1999; Grigos, 2009; Grigos and Patel, 2007). Conture and colleagues have used the observation that CWS differ from CWNS in specific temperament constructs to propose that for these children, normal, and in some cases, subtly deficient speech and language development may interact with environmental factors and the child's reaction to this external and internal variable in such a way that stuttering is exacerbated. For example, if a CWS possesses a vulnerable temperament, he or she may react to disruptions in speech and make associations between particular speech mistakes and emotional reactions to them, which may maintain or exacerbate stuttering. Conture et al. stressed that variable, situationally driven emotional behaviors (i.e. related to environmental context), as opposed to stable aspects of temperament, are what influence the child's proclivity to maintain or exacerbate stuttering. In particular, both Zebrowski (1994) and Anderson et al., (2003) have suggested that CWS who are less easily distracted "may be less likely to allow external stimulation to divert their attention from disruptions or mistakes in their own speech (p. 1229)", thus leading to a tendency to "stay longer", struggle, or increase physical tension during an instance of speech disfluency or speech error (Anderson, 2003 p.1229).

1.5. Purpose

Following the combined observations made by Zebrowski (1994), Anderson et al. (2003), and predictions based on the Communication-Emotional model of stuttering (Conture et al., 2006), we speculate that some CWS may show a relationship between their ability to regulate their attention and adapt to new situations and the production of longer sound prolongations in conversational speech. In addition, we predicted that those CWS who produced more and longer sound prolongations would also show slower articulation rate in conversational speech. Presently, there are no published data showing that there is such a relationship for preschool CWS.

As a first step, we examined the relationships between articulation rate and both overall frequency of SLDs and sound prolongation, and between articulation rate and the duration of individual types of SLDs (i.e. sound prolongations, sound-syllable repetitions, monosyllabic whole-word repetitions and disfluency (SLD) clusters. Second, we analyzed the relationship between specific temperament constructs that have previously been shown to distinguish CWS from their normally fluent peers, and the duration of sound prolongations. Finally, we included two measures of articulation rate (syllables and phones per second) to attempt to replicate and perhaps extend previous research that they are distinct. Our long term goal is to determine whether these variables, and their relationships, can serve to subtype CWS with regard to the developmental pathways of early stuttering.

2. Method

2.1. Participants

Nineteen children who stutter (14 boys and 5 girls; mean age = $3:9$; range = $2:10 - 5:10$ (years: months) participated in the study. Participants averaged 14 months post onset of stuttering, with a range from 2–36 months post-onset. Children were recruited via advertisements in local newspapers and referrals from speech language pathologists throughout the states of Iowa and Illinois. Participants for this study were part of a larger multi-site study conducted at the University of Illinois, University of Iowa and University of Wisconsin at Milwaukee and directed by investigators at the University of Illinois (E. Yairi and N. Ambrose, RO1-DC05210). All children were native speakers of American English with no history of neurological, hearing, or intellectual problems. Children were considered to be stuttering if they met two standard criteria (Zebrowski, 1994; and others): (a) they produced 3 or more stuttering-like disfluencies (SLD) (i.e. sound-syllable repetitions, sound prolongations, monosyllabic whole word repetitions) per 100 words of conversational speech; (b) their parents believed that the child stuttered. Once a child qualified for the study, a speech-language pathologist assessed the severity of stuttering based on the stuttering severity scale devised at the University of Illinois (for detailed description, see Yairi & Ambrose, 1999). This is a seven point scale in which zero indicates "normal" and 7 "very severe" stuttering. Severity ratings for children in the present study ranged from 1.33 to 5.50. Participant age, sex, post-onset interval, number of SLDs and severity ratings are presented in Table 1.

2.3. Procedures and Measures

2.3.1. Speech samples—Conversational speech samples were obtained from each child as she or he played with a parent using Play-Doh. All parent-child interactions took place at a child-sized table, and parents were instructed to play with their child as they might at home. Each parent-child interaction was video recorded using a stationary video camera (Sony, model DCR VX 2000) placed approximately 1 meter from the child. The audio signal was obtained using a lapel microphone (Shure MX183BP) that was placed approximately 20 cm from the child's lips, and a table top microphone (Shure MX3930) that was placed on the table. The audio signal was separately recorded on a high quality CD recorder (HHB CDR830). These CDs were used for subsequent analysis of the child's speech.

2.3.2. Transcription of conversation samples—One contiguous speech sample from each CWS was transcribed for analysis. The total number of words that could be analyzed in individual speech samples varied across children, ranging from 169 to 343 words, with an average of 294 words. All speech samples were orthographically and phonetically transcribed using PEPPER software (Programs to examine phonetic and phonologic evaluation records) developed by Shriberg (1986). PEPPER is a software program for

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analysis of continuous speech samples. The program accommodates for narrow and broad phonetic transcription using the International Phonetic Alphabet (IPA). PEPPER allows for several phonologic analyses, including: intended and realized words, phonemes, features, item, percent consonants correct, and natural process analysis. For the purposes of the present study, PEPPER was used to transcribe each of the child's utterances within the sample in orthographic form. Both the ideal or adult form and the actual form for both grammar and phonology were coded. The completed PEPPER transcripts served two main purposes. First, they were used to guide the examiner during visual and auditory inspection of the spectrograms for the identification of disfluencies. Second, they provided the realized phonological transcription of each utterance which was then used to count the number of phones and syllables within each utterance necessary for the measure of articulation rate.

2.3.3. Measures of SLD type, frequency and duration—Two frequency measures were obtained from each sample: the number of all stuttering-like disfluencies (SLDs; Yairi & Ambrose, 1999) in the sample, as well as the number of each type of SLD produced by the child. In addition, the sound prolongation index, or SPI (proportion of the total number of SLDs that are sound prolongations; Schwartz &Conture, 1988; Zebrowski, 1994) was calculated for each child's sample. Stuttering-like disfluencies included sound-syllable repetitions, monosyllabic whole-word repetitions sound prolongations (audible and inaudible), and SLD clusters (i.e., conjoined prolongations and repetitions of the same sound or syllable; LaSalle & Conture, 1995). Judgments of disfluency type were made using a classification scheme derived from those described by Conture (1990) and Yairi & Ambrose (1992) that have yielded a high degree of intra- and interjudge reliability across a large number of studies (e.g. Kelly & Conture, 1992; Yairi & Ambrose, 1992; 1999; Zebrowski, 1991, 1994).

Measures of SLD duration were made acoustically using *PRAAT*: Doing Phonetics by Computer (version 5.0.01) (Boersma & Weenink, 2007), and based on guidelines established in prior acoustic studies of stuttering duration (e.g. Kelly & Conture, 1992; Zebrowski, 1991; 1994). Using both speech playback and the sound waveform, the speech segment of interest (i.e. SLD or entire utterance) was selected and a Fast-Fouriertransformation based spectrogram was displayed. Duration of sound-syllable and monosyllabic whole-word repetitions, and SLD clusters, was measured from the onset of acoustic energy associated with the disfluent initial sound in a word until the termination of acoustic energy for the final iteration of the repeated sound or syllable. For audible sound prolongations, duration was measured from the onset of acoustic energy associated with the disfluent sound in a word to the termination of acoustic energy for the prolonged sound. The duration of inaudible sound prolongations was measured from the cessation of acoustic energy associated with the preceding sound to the onset of the fluent sound that followed. Inaudible sound prolongations at the beginning of utterances were excluded from the analysis.

2.3.4. Measure of articulation rate—The articulation rate of each child's speech sample was obtained in both syllables and phones per second, using PRAAT, and following previously established guidelines (e.g. Hall et al., 1999; Kelly & Conture, 1992; Kelly, 1994; Sawyer, Chon, & Ambrose, 2008; Zebrowski, 1991, 1994). Mean articulation rate was obtained by dividing the total number of fluent syllables and phones in each utterance by the total duration of the utterance, following the subtraction of the duration of all disfluently produced syllables and phones, as well as all within-utterance pauses of 250 ms or longer. This method has been widely used in studies of articulation rate in both typically developing and stuttering children (e.g. Kelly & Conture, 1992; Kelly, 1994; Miller, Grosjean & Lomanto, 1984; Sawyer et al., 2008; Walker, Archibald, Cherniak & Fish, 1992; Yaruss, 1997; Yaruss & Conture, 1995;1996; Zebrowski, 1994). Utterance duration was measured in

milliseconds by placing cursors at the onset and offset of the utterance of interest as displayed on a wide-band spectrogram. Onset was defined as the first burst of acoustic energy associated with the production of a particular phoneme on the corresponding spectrogram, and offset was defined as the last visible peak of acoustic energy on the corresponding spectrogram. In cases when stop sounds started or finished the utterance the onset and offset of the sound was defined as the visible burst of energy associated with the release of the stop sound.

Two important measurement procedures should be noted here. First, we decided to measure articulation rate across each child's entire sample, as opposed to using only perceptually fluent utterances, as has been done elsewhere (e.g. Hall et al., 1999). This decision was based on our desire to maximize the size of usable speech samples, but also because using contiguous utterances within a single sample provides an ecologically valid context for exploring potential relationships between rate and stuttering. That is, given previous suggestions that there may be an "overflow" effect (Conture, 1990; Zebrowski, 1994) present when a stuttered disruption is produced, we wanted to ensure that any potential anticipatory or carry-over temporal effects (e.g. Viswanath, 1989) on the surrounding fluent syllables and words would be captured and reflected in the articulation rate of both the utterance and the sample as a whole. In addition, as previously stated the method we used has wide precedence in the literature, and therefore provides the opportunity to compare our results with a relatively large number of previous studies. Second, we decided to measure rate in both phones and syllables per second to examine the degree to which they were (or were not) correlated, thus allowing us to makes some conclusions about differences in their potential for identifying subtypes of stuttering development (e.g. Hall et al., 1999).

2.3.5. Assessment of temperament—A temperament profile of each child was obtained by using the short form of the Children's Behavior Questionnaire (CBQ) (Putnam & Rothbart, 2006; Rothbart et al., 2001). The CBQ is a well-established assessment of temperament in children from three to eight years of age, based on parent report. The short form of the *CBQ* consists of 94 items across 15 scales that comprise temperament. These include: Activity Level, Anger/Frustration, Approach/Positive Anticipation, Attentional Focusing, Discomfort, Falling Reactivity/Soothability, Fear, High Intensity Pleasure, Impulsivity, Inhibitory Control, Low Intensity Pleasure, Perceptual Sensitivity, Sadness, Shyness, Smiling and Laughter.

Scores from each of the 15 scales are collapsed into three broad constructs or factors that characterize temperament. Fifteen temperamental characteristics (scales) form three broad dimensions, or factors, of temperament: Surgency/Extraversion, Negative Affectivity, and Effortful Control. Internal consistency for the scales has been established with data from 590 predominantly Caucasian children of middle socioeconomic status (Cronbach's αs range from .65 to .93; Putnam & Rothbart, 2006). Moreover, the validity of the CBQ has been recognized through numerous studies of child temperament (for review see Putnam & Rothbart, 2006).

Parents in the present study filled out the CBQ questionnaire during their initial visit. They were instructed to respond to each item to the best of their ability; no other specific guidelines were provided. The questionnaire uses a seven point scale to describe the frequency with which parents observe a certain behavior in their child, with 1 being "extremely true" and 7 "extremely untrue." Parents also have a choice of "not applicable" for each item. Examples of items include "can wait before entering into new activities if he/ she is asked to", "is good at following instructions," and "can easily stop an activity when he/she is told 'no' from the Inhibitory Control scale, and " when practicing an activity, has a hard time keeping her/his mind on it", "when drawing or coloring in a book, shows strong

concentration," and "sometimes becomes absorbed in a picture book and looks at it for a long time" from Attention Focusing.

For the purposes of this study, scores from the individual Inhibitory Control and Attentional Focusing scale were chosen for correlation analysis. According to Putnam (2008, personal communication), of the constructs assessed through the CBQ, these best represent a child's ability to adapt to novel circumstances and maintain attention on a particular task, both characteristics that were shown to differentiate CWS from CWNS in the study by Anderson et al. (2003). In addition, these skills are prominent factors in the Communication-Emotional model of stuttering development, and central to prior speculations that difficulty in shifting attentional focus and adapting to novel circumstances may relate to increased frequency and duration of prolonged sounds and decreased articulation rate, (i.e. Anderson et al., 2003; Zebrowski, 1994).

2.4. Reliability for measures of speech disfluency and articulation rate

The initial segmentations and measures were performed by the first author. To ensure intrajudge and interjudge reliability an entire speech sample (300-word) from one participant was chosen at random for re-evaluation by the first author and another examiner, who was experienced in acoustic analysis. We separately assessed reliability of (1) segmentation accuracy (identification of SLDs, other disfluencies, and fluent speech intervals) (2) acoustic measurements of duration of fluent speech intervals and SLDs.

Percent agreement (agreements/(agreements + disagreements)*100) (Hunt, 1986) was calculated to estimate reliability of examiners segmentation accuracy. All SLDs and fluent speech intervals were identified and marked on the transcripts by each examiner independently. Point-by-point comparison of transcripts was completed. The criterion for an agreement between the two transcripts was an exact match. For intra-judge reliability the time between the first and the second examination of the transcript was about 6 months. Both intra-judge and inter-judge agreement based on re-segmentation of an entire speech sample from one participant was 91%. Chance-correction of the intra-judge and inter-judge agreement using Cohen's Kappa (Hunt, 1986) or a similar statistic was not possible because segmentation of speech samples into fluent intervals and disfluencies of different types does not involve closed set of judgments.

Pearson correlation coefficients were calculated to evaluate reliability of durational measures between and within the examiners. Intra-judge reliability based on re-measurement of 114 speech intervals (the time between the measurements was about 6 months) was 93%. Inter-judge reliability based on re-measurement of 95 speech intervals was 83%.

3. Results

3.1. Frequency and duration of SLDs in CWS

The 19 CWS produced a total of 91 sound-syllable repetitions, 149 sound prolongations, 96 monosyllabic whole-word repetitions, and 13 disfluency clusters. The Sound Prolongation Index (SPI) was calculated for each participant by diving the number of prolongations produced in the sample by the total number of SLDs. Mean SPI for the 19 participants in the study was 0.48 percent (SD=0.18; range=0.07–0.79).

As Table 3 shows, the mean duration of sound-syllable repetition was 0.87 seconds (SD=0.38 sec; range=0.28–1.69 sec), and the average duration of monosyllabic whole-word repetitions was 0.99 seconds (SD=0.25 sec; range=0.65–1.63 sec). Mean duration of audible and inaudible sound prolongations combined was 0.63 seconds (SD=0.21 sec; range=0.37– 1.09s). Finally, the mean duration of disfluency clusters was 1.78 seconds (SD=0.97s;

range=0.78–3.59s). The average duration and range of SLDs produced by the children in this study was generally consistent with prior reports for same-aged CWS (Kelly & Conture, 1992;Louko, Edwards & Conture, 1990;Zebrowski, 1991,1994). Individual data for each participant is presented in Table 2, and descriptive analysis is presented in Table 3.

3.2. Articulation rate

Recall that two measures of articulation rate were obtained; syllables and phones per second. The average articulation rate in syllables per second was 2.90 (SD=0.60; range=1.83–4.02 syllables per second), while the average rate measured in phones per second was 6.98 (SD= 1.48; range= 4.42–9.58 phones per second). Analysis of a relationship between age and articulation rate indicated a significant positive correlation ($r=0.557$; $p<0.01$), as was expected; that is the older CWS produced more syllables per second in their conversational speech. These means are slower than what has been reported in previous work; however, when considering both the standard deviation and the range of articulation rate observed in the present study, our results appear to be generally comparable to those from earlier studies that used similar acoustic analysis methods for the speech of young CWS. For example, Kelly and Conture (1992) reported articulation rate in CWS aged 3:3 to 4:8 (years: months) to be 200.21 syllables per minute, which corresponds to 3.34 syllables per second; Kelly (1994) observed that articulation rate in CWS in her study was 197.2 syllables per minute (3.29 syllables per second), although the age range of children in their study (2:7 to 10:1; years: months) was much greater than in the present study.

In a study that measured articulation rate in utterances with no pauses or disfluencies, Hall et al. (1999) reported articulation rate measured in CWS (39–55 months, which is comparable to the children in the present study) over three separate visits (initial visit, one-year follow up visit and two-year follow up visit). At the initial visit, Hall et al., reported a mean articulation rate of 3.18 syllables per second or 7.68 phones per second. Using a similar method, Sawyer et al. (2008) measured mean articulation rate for preschool CWS (mean age 3:4; years: months) to be 3.47 syllables per second. For three and five year-old CWNS, Walker et al. (1992) reported mean articulation rate to be 3.82 syllables per second (8.42 phones per second) and 4.28 syllables per second (9.47 phones per second) respectively. In a different study, Walker and Archibald (2006) studied the articulation rate of four year-old CWNS and reported it to be 3.56 syllables per second. The most likely explanation for the small differences in findings between and among all of these studies, including our own, is the differences in subject samples (e.g. mean and range of ages, etc.) and methodology (e.g. rate measured manually versus acoustic analysis; type of speech sample analyzed).

As previously discussed, Hall et al. (1999) concluded from their study that articulation rate measured in phones per second, as opposed to syllables per second, was related to different pathways of stuttering development (i.e., persistence versus recovery). While that may be the case when comparing subtypes, our findings showed that within the group of CWS these two measures were significantly, and strongly correlated in conversational speech (r=0.98; p<0.0001). This observation, along with similar findings by Walker et al. (1992) for sameaged CWNS suggests that for general use, and particularly for the purposes of our study, phones and syllables per second are equivalent measures of articulation rate.

3.3. Temperament

Recall that a primary purpose of the present study was to observe whether a significant relationship exists between CWS ability to regulate their attention and adapt to new situations and the production of more and longer sound prolongations in their conversational speech. In order to examine whether such a relationship exists, we calculated the group means for the three main factors in the CBQ, using the scores from the 15 individual scales

(including Inhibitory Control and Attentional Focusing). Because these two scales best reflect the two characteristics that served to distinguish CWS from CWNS in the study by Anderson et al. (2003), we chose to use them in the correlation analysis as well.

The mean score for Surgency/Extraversion was 4.70 (SD=0.55; range=3.52–5.91), the mean score for Negative Affectivity was 4.06 (SD=0.86; range=1.82–5.75), and the mean score for Effortful Control was 5.10 (SD=0.62; range=3.94–6.13). Mean scores for Inhibitory Control and Attentional Focusing were 4.36 (SD=0.91; range=2.67–5.83) and 4.88 (SD=1.11; range=2.67–6.83) respectively. Pearson correlation analysis between sound prolongation duration and the three broad dimensions of temperament comprised (i.e. Surgency/Extraversion, Negative Affectivity, and Effortful Control), and Inhibitory Control and Attentional Focusing scales, revealed no significant correlations between sound prolongation duration and any of the three factors or the two scales of interest from the CBQ (see Table 5).

While not a purpose of this investigation, we were interested to know how the CWS in our study compared to same aged typically developing children on the two scales of interest; Inhibitory Control and Attentional Focusing. Mean scores on the CBQ for typically developing children whose parents completed the CBQ were reported by Rothbart et al., 2001, and we used those scores for our comparison. According to Rothbart et al., the mean score for Inhibitory Control for typically developing three year-olds was 4.26 (SD=0.73; range=2.46–6.80) and 4.41 (SD=0.68; range=2.93–6.25) for Attentional Focusing. Three year-old CWS in our study had a mean score of 4.29 for Inhibitory Control and 4.58 for Attentional Focusing. For typically developing four and five year-olds the mean score for Inhibitory Control was 4.75 (SD=0.83; range=1.62–6.92), and 4.50 for Attentional Focusing (SD=0.68; range=2.5–6.63). Four year-old CWS in our study had a mean score of 4.48 for Inhibitory Control and 5.38 for Attentional Focusing. Individual data for both scales are presented in Table 6.

3.4. Relationship between frequency and duration of SLD, and articulation rate

To examine the relationship between the duration of SLDs, frequency of sound prolongation, and articulation rate in the conversational speech of CWS, Pearson correlation coefficients were calculated between articulation rate and measures of SLDs. Analyses revealed a significant negative correlation between (1) articulation rate and average duration of sound prolongations (r= −0.584; p< 0.01), and between articulation rate and overall frequency of SLDs in the conversational samples ($r = -0.488$; $p < 0.05$). That is, children, who produced more SLDs in their speech, and children who produced longer sound prolongations, produced slower articulation rates. There were no significant relationships between frequency of individual SLDs (e.g. SPI) and articulation rate, or between articulation rate and the duration of sound-syllable repetitions, whole-word repetitions, or SLD clusters. Table 4 presents Pearson correlation coefficients between articulation rate in fluent speech and measures of disfluencies in speech of CWS.

4. Discussion

This study yielded three main findings. First, our prediction of a correlation between the ability to regulate attention and adapt to new situations, and the duration of sound prolongation was not born out. Second, those CWS with a relatively higher frequency of SLDs overall produced slower articulation rate, but there was no significant relationship between articulation rate and the frequency of sound prolongations as reflected in the SPI. Finally, there was a significant negative correlation between articulation rate and the duration of sound prolongation, but not with the duration of any other SLD; those CWS who

produced a slower articulation rate also produced longer sound prolongations when compared to their peers whose articulation rate was faster.

The observation that there was no significant relationship between parent-judged inhibitory control or attentional focusing and the duration of sound prolongation to some extent contradicts the speculations by Anderson, et al., 2003; Schwenk et al., 2007; Conture, 1990; Conture et al., 2006 and Zebrowski, 1994, among others, that specific aspects of a child's temperament are proximally related to specific features of stuttering. That is, if temperament is a significant factor in the development of stuttering, it does not manifest in the (local) behavioral characteristics of the child's stuttering, but rather contributes more globally to the way a child reacts to his or her stuttering. That being said, the present findings provide some direction for future studies that attempt to evaluate models of childhood stuttering that incorporate temperament, such as the CE model described earlier. For example, rather than motivating reactions at the level of speech physiology, increased reactivity to stuttering and less ability to regulate attention may lead a child to avoid specific speaking or social situations, and ultimately particular sounds and words and even day-to-day experiences or life choices. There are numerous anecdotal accounts from the clinical literature of these reactions to stuttering, and how they influence treatment outcome.

Assuming that temperament may be more likely to play a role in the child's experience, and not necessarily production, of stuttering, future research should examine the relationship of temperament to a child's awareness of stuttering and his or her thoughts, attitudes and beliefs about talking and stuttering. For example, can specific measures of temperament predict a child's score on such instruments as the Communication Attitude Test – Revised (De Nil & Brutten, 1991), or the Kiddy CAT (Vanryckeghem & Brutten, 2007) for younger children? Similarly, is there a relationship between a preschool child's temperament and treatment outcome in (quasi) standardized programs for stuttering, such as Lidcombe (yr) or Parent-Child Interaction Therapy (PCIT) (Millard, Nicholas, & Cook, 2008).

There is evidence that speech planning and production are influenced by processes that, according to Maner, Smith and Grayson (2000) "are considered to be relatively remote from the motor output stage" (p. 571). Such processes may include temperament. Presently, existing studies in the temperament of CWS, including the present one, have examined only the surface aspects of temperament, and these observations alone are not sufficiently sensitive to the ways in which associated levels of autonomic nervous system reactivity are related to stuttering. The challenge, then, is to develop ways to reliably observe the link between the behaviors of temperament, their autonomic nervous system correlates, and the relationships between these factors and speech fluency in children. One possible framework for this analysis can be taken from the work of Alm (2004a, b), and earlier by Peters and Guitar (1991). These authors have proposed that on a speech motor level, changes in reactivity of the autonomic nervous system can lead to a "freezing response" that is manifested in muscle activation and articulator movement. Alm (2005) has gone so far as to speculate that repetitions and prolongations of sounds and syllables are the result of a complex interaction between increased activation of motor, emotion and cognitive factors (so called "positive symptoms") and the absence of typical functioning within and across these domains (referred to as "negative symptoms") in individuals who stutter. Results from the present study, and prior work, are of course unable to support or refute this view, but through the use of different paradigms (see Future Research) this relationship may be explored.

The second main finding in the present study was the significant negative correlation between articulation rate and the frequency of SLDs in conversational speech. This observation is similar to that of Zebrowski (1994) for school-age CWS; however, unlike

Zebrowski, we did not find a significant relationship between articulation rate and the frequency of sound prolongation (i.e. SPI). One interpretation of this latter discrepancy may lie in the developmental aspects of stuttering; specifically, an increase in prolonged sounds relative to other SLDs may either predict or co-occur with more chronic forms of the disorder, particularly in children who persist beyond the period when unassisted recovery seems likely (compare Throneburg and Yairi (2006) for dysrhytmic phonation).

Finally, there was a significant negative correlation between articulation rate and the duration of sound prolongation, but not with the duration of any other SLD; those CWS who produced a slower articulation rate also produced longer sound prolongations when compared to their peers whose articulation rate was faster. This observation, again, is similar to that of Zebrowski (1994) for school-aged children. The finding of this relationship in the speech of younger children who are closer to the onset of stuttering, in the absence of an observed relationship between temperament and sound prolongation duration, can be interpreted in different ways. First, it may be the case that the way we chose to measure articulation rate, by subtracting disfluencies and pauses to yield fluent utterances, in part set the stage for such a relationship to emerge. This might be explained by considering the findings from studies of speech physiology that have shown that the fluent speech surrounding instances of stuttering is different from fluency that is not in the vicinity of stuttered disruptions (e.g. Viswanath, 1989). That is, although the instances of disfluency and pauses were removed for the present measures of articulation rate, "anticipatory" "carryover" or "overflow" (Zebrowski, 1994) effects in the syllables or words either preceding or following the stuttered disruption remained. If such an effect is characterized by increased movement, phone, or syllable duration, the result would be a decrease in articulation rate. And, this effect becomes more apparent as the number of SLDs in general increases.

Of interest is that in the present study, this relationship was significant for articulation rate and sound prolongation duration alone (see Table 4), despite the fact that the mean durations for all other SLDs were longer than that for sound prolongation (see p. 16, Results). It seems reasonable to conclude from these data that while prolonged sounds are less temporally salient, or disruptive in the flow of continuous speech than other types of stuttering, they have a greater influence on the surrounding fluency. The implication here is that prolonged sounds, as opposed to other types of SLD, may have a motor consequence that can become habituated over time. While such "motor consequences" are unknown, perhaps findings from studies that have shown speaking rate effects on articulator movement, segment duration, and both spatial and temporal variability in both within stuttering and nonstuttering speakers are relevant (e.g. Adams, Weismer,& Kent, 1993; Kleinow, Smith and Ramig, 2001; Sim and Zebrowski, 1996). Among other things, these studies have shown that speech gestures produced at habitual or fast speaking rates involve unitary movements, whereas gestures produced at slow speaking rates consist of multiple movements. In addition, compared to habitual and fast speech rates, a slower speech rate is associated with greater variability in articulator movement and segment duration. Within the stuttering literature, researchers have long speculated that increased variability in both temporal and spatial aspects of speech production reflect instability in the system and therefore increased vulnerability to speech disruption (e.g. Smith and Kleinow, 2000).

Our findings along with evidence from different lines of theoretical, clinical, and experimental research in stuttering and normal speech can be interpreted to suggest that sound prolongations, across a range of durations, may represent a distinct type of SLD, not just in their obvious perceptual characteristics, but in their potential influence on overall speech production at multiple levels. Of course, the opposite is also likely, and in fact has been most widely discussed in the literature. That is, that people who stutter use different motor control strategies (e.g. "feedback" versus "feed forward"; Max, Guenther, Gracco,

Ghosh and Wallace, 2004), exhibit highly variable and unstable speech production systems (e.g. Smith & Kleinow, 2000, and others), and deficient phonological encoding (e.g. Postma & Kolk, 1993), all leading to various forms of stuttered disruption.

5. Future Research

As previously discussed, additional research in the temperament of CWS should explore new avenues to determine whether temperament as mediated by the autonomic nervous system is simply a covariate, or is in fact related to some aspect of stuttering development, behavior or experience. That is, future experimental work should move toward measurement, at multiple levels, of potential relationships between both observable and unobservable (i.e. autonomic nervous system) features of temperament and their relationship to speech production in children who stutter. As one example, consider the work of Wolfe and Bell (2004; 2007), who examined the relationship between temperament, physiological functioning, and working memory in a group of four year-old typically developing children. In this study, relations between children's scores on the effortful control scale of the CBQ, the cognitive processes of working memory and inhibitory control (as assessed by a version of the Stroop test), and two physiological measures – heart period and cortical responsewere correlated. Findings revealed a significant relationship between temperament, physiological response, and task performance. Similarly, Burgess, Marshall, Rubin and Fox (2003) observed significant associations between temperament (i.e. inhibited versus uninhibited,) cardiac measures (heart rate and respiratory sinus arrhythmia), and social behavior in children. Further, using a longitudinal design, these researchers observed that the interaction of child temperament, parent-child interaction, and cardiac behavior in infancy is predictive of later social behavior. The results of these and other studies lend support to biopsychosocial models of development that seem to fit well with current theories of how stuttering emerges in young children (e.g. Smith and Kelly, 1997). As such, they can provide a framework for similar investigations in childhood stuttering that attempt to link temperament to autonomic nervous system function and salient measures of speech production (i.e. articulator movement, laryngeal behavior, articulation rate, and fluency. Added to this, the experimental paradigm used by Weber and Smith (1990) to show a positive relationship between sympathetic arousal and severity of stuttering in adults can be modified for children, and include measures of temperament.

Finally, the determination of which, if any, temperamental constructs are associated with the attitudes or emotions experienced by CWS, as opposed to the behavior of stuttering, has implications for both the development and treatment. Similarly, the concept that prolonged sounds reflect a distinct subtype of speech disfluency influencing stuttering development, and affecting multiple levels of speech fluency (e.g. articulation rate, speech and language planning) warrants attention.

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Table 1

Articulation rate, measures of stuttering-like disfluencies duration and frequency, SPI, number of words analyzed for 19 children who stutter. Articulation rate, measures of stuttering-like disfluencies duration and frequency, SPI, number of words analyzed for 19 children who stutter.

Articulation Rate and Disfluency Profile of Participants

Pearson Correlation Coefficient Matrix for Articulation Rate(phones per second) Pearson Correlation Coefficient Matrix for Articulation Rate(phones per second)

Pearson Correlation Coefficient Matrix for Average Duration of Prolongations Pearson Correlation Coefficient Matrix for Average Duration of Prolongations

CBQ data

Note. Norms for 3 year-olds are 4.41 for attention focusing score and 4.26 for inhibitory control score; for 4 and 5 year-olds the norms are 4.5 for attention focusing score and 4.75 for inhibitory control score