Speech Production Analyses:

Characterizing Typical Development versus Childhood Apraxia of Speech

by

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Childhood Apraxia of Speech (CAS) is a controversial diagnosis that is frequently based on clinical perceptions, with treatment success based on the same perceptual measures. Because additional, less abstract diagnostic and treatment outcome procedures are needed, a series of quantitative trials were chosen for the current research to potentially assist in the diagnosis and evaluation of treatment for those with CAS. The study included two participants, one child with typically developing (TD) speech and one diagnosed and verified by current protocols as having CAS. Kinematic and acoustic measures were used to calculate spatiotemporal index, speech-pause time, and lexical stress. The spatiotemporal index was factored using kinematic data and computer-based algorithms. Acoustic data were used to evaluate speech versus pause time as well as lexical stress. Speech-pause time was calculated by measuring speech time in comparison with pause times both between and within words. Lexical stress was calculated by computing ratios involving vowel length, mean frequency, and mean amplitude of the first syllable over the second syllable.

The participant with CAS displayed greater inconsistency with both the lower and upper lip during repetitions of “Buy Bobby a puppy;” with a higher factored spatiotemporal index for

both lips as compared to that of the TD participant. In a story retell task, acoustic analyses of participants' responses revealed increased total utterance time in addition to increased pause time percentage in the participant with CAS versus the TD participant. During repetition of eight trochaic words, the participant with CAS presented greater mean lexical stress while the TD participant displayed stress primarily on the initial syllable. These results provide feasibility for using the given measures to differentiate speech productions of TD children from those with CAS. Additional study of the current measures on a larger scale with TD speech participants as well as in comparison with participants exhibiting other speech sound disorders is recommended.

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Characterizing Typical Development versus Childhood Apraxia of Speech

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by

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Chapter I: Review of Literature

Although historically controversial, the definition of Childhood Apraxia of Speech (CAS) recently has been identified among clinicians as a neurologically based, motor-planning speech sound production disorder (American Speech-Language-Hearing Association [ASHA], 2007). Exactly how and what differentiates the speech production of children with CAS from typically developing (TD) children has yet to be determined (ASHA, 2007; Shriberg, Aram, & Kwiatkowski, 1997a). Currently, CAS is regarded as "a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (ASHA, 2007, p. 2)." Although most professionals agree that CAS is a disorder based in motor planning, little is known regarding the origin and nature of CAS (Shriberg et al., 1997a). Due to the lack of definitive data available regarding the disorder, no standardized measures have proven effective in identifying children with CAS and monitoring change with treatment. Motion analysis of speech movements, however, has yet to be investigated as a possible procedure to aid in such management of CAS.

Kinematics of Speech and Non-Speech Movements

While early kinematic modeling of speech movement was measured using cantilever beams and strain gauges (Caligiuri, 1987), recent studies have progressed to computerized tracking and kinematic analysis of movement (Green et al., 2002; Jiang, Alwan, Bernstein, Keating, & Auer, 2000; Jiang et al., 2001; Trotman et al., 1998). As mentioned, motion analysis has been used to obtain information on the variability in timing and/or amplitude of the speech signal (Jiang et al., 2000; Jiang et al., 2001; Löfqvist & Gracco, 1997; Smith et al., 1995; Smith & Goffman, 1998; Trotman et al., 1998), the creation of human face models (Lucero, Maciel, Johns, & Munhall, 2005), the development of stability and coordination in speech (Green,

Moore, Higashikawa, & Steeve, 2000; Green et al., 2002), as well as differences between typical and atypical speech productions (Caligiuri, 1987; Caruso et al., 1988; McClean et al., 1990).

Löfqvist and Gracco (1997) explored the force and variability of bilabial stop production in TD adults. Findings indicated that during the process of bilabial stop production, TD adults showed negative aperture of the upper lip over the lower lip due to high velocity of lip motion directly before closure. Trotman et al. (1998) focused on motion analysis of non-speech movements such as smiles, cheek puffs, and lip pursing. Jiang et al. (2000) conducted a study to determine the effects of facial movements, tongue movements, and acoustics of speech. This study utilized motion analysis and an Electo-Magnetic Midsaggital Articulometer (EMMA) to evaluate tongue movement and acoustics of speech. Jiang et al. (2001) again used motion analysis to measure TD adult lip aperture in regards to lip-reading intelligibility. The study investigated many other related factors such as acoustic information, lip excursion, and lip height. Lucero et al. (2005) developed a model of the TD adult human face using kinematic data. The study integrated use of 38 reflective facial markers, which were subsequently grouped into clusters that served as a connected region showing similar movement patterns during speech.

Kinematic data also has been used to map the development of speech in children when compared to adults. Green et al. (2000) researched development of lip and jaw coordination during the early years of speech development. The results showed a significant coordination shift during the first few years of life, becoming more refined by age 6. However, there was a continual refinement after the age of 6, leading to differences between data collected with 6-year-old children and TD adults. Green et al. (2002) researched the development of jaw and lip control in speech production. Although they found differences between 1-year, 2-year, and 6-year old children in jaw and lip control, variation within age groups was minimal.

Kinematic information has been utilized to typify certain patterns associated with particular speech disorders (Caligiuri, 1987; Caruso et al., 1988; McClean et al., 1990). Caligiuri (1987) measured the labial rigidity of participants with Parkinson's disease. It was found that lip rigidity negatively influenced the amount of displacement of a person with Parkinson's versus that of an unaffected adult. Kinematic analysis also has been conducted on individuals who stutter (Caruso et al., 1988; McClean et al., 1990; Smith & Kleinow, 2000). Caruso et al. (1988) studied lip and jaw coordination patterns of non-stuttered speech in individuals who stuttered and compared these data to speech productions of TD adults. Differences were documented specifically in sequencing of movements (Caruso et al., 1988). McClean et al. (1990) conducted a similar study on lip closure among individuals who stuttered versus TD peers. However, they found minimal kinematic differences between the two groups. Smith and Kleinow (2000) also found subtle and inconsistent kinematic differences between TD participants and those who stuttered.

The previously mentioned analyses have been performed using kinematic data including basic measurements of velocity, aperture, and amount of movement during speech and non-speech productions (Caligiuri, 1987; Caruso et al., 1988; Green et al., 2002; Jiang et al., 2001; Löfqvist & Gracco, 1997; McClean et al., 1990; Trotman et al., 1998). Another measure which can be calculated utilizing kinematic data from repeated stimuli is known as the Spatiotemporal Index (STI) (Smith, Goffman, Zelaznik, Ying, & McGillem, 1995). The STI is calculated using the sum of lip excursion standard deviations at set intervals across the whole signal. The analyses are overlapped and normalized in order to observe the consistency and stability of the signal. As typical speech is a highly precise and repetitive motor task, it is believed to show high stability and consistency on repeated utterances. Thus, lower variability in speech production on repeated

phrases would be reflected in a low STI score. While utilizing these measures, Smith et al. (1995) found that speech rates have a significant impact on participant's STI. Although no differences were found between regular and accelerated rates, a slow rate resulted in significantly higher STI values. Using similar techniques, Wohlert and Smith (1998) evaluated the impact of age on STI values. Findings suggested that older adults had more variability than younger adults for all measured speaking rates during utterance repetitions. However, differences in degree of lip aperture and habitual rate were found to be lower in older adults, which may have contributed to the higher STI results. Research also has indicated a progression in development of stability with age (Maner, Smith, & Grayson, 2000; Smith & Goffman, 1998). Both studies found that younger children have consistently higher STI values than their adult counterparts. Smith and Goffman (1998) noted specifically that by age 7, children had reached STI values similar to a typical adult. Maner, Smith, and Grayson (2000) also researched the impact of surrounding speech productions, finding that STI was significantly increased when a target phrase was produced in a complex sentence versus in isolation.

Childhood Apraxia of Speech

Information on CAS has become more available as researchers have attempted to present diagnostic criteria for differential diagnosis (ASHA, 2007). However, the research has led to minimal empirical data that can be used to diagnose the disorder (Shriberg, Aram, & Kwiatkowski, 1997b). The main areas of focus, however, have centered on perceptual speech characteristics, acoustic and/or prosodic characteristics, and genetics (ASHA, 2007). Currently, non-speech motor difficulties frequently are being used to distinguish CAS from dysarthria using formal examinations such as the *Verbal Motor Production Assessment for Children* (Hayden & Square, 1999).

Several studies have identified perceptual speech characteristics that are more likely observed among children diagnosed with CAS. These conclusions are based on clinical observations and evaluations of TD children, children with developmental speech sound disorders, as well as children with suspected CAS (Davis, Jakielski, & Marquardt, 1998; McCabe, Rosenthal, & McLeod, 1998). Both Davis et al. (1998) and McCabe et al. (1998) indicated that the following characteristics may possibly be considered markers for CAS: inconsistency in speech errors; vowel errors; increase in errors relative to length and complexity of the utterance; groping; and variable ordering of sounds, words, or parts of words. Concerns with these results, however, are based on the fact that some of the noted characteristics also are observed among children not diagnosed with CAS (McCabe et al., 1998; Davis et al., 1998). Speech motor difficulties, mainly with repetition of syllables (e.g., /bababa/) and alternation of syllables (e.g., /pataka/) known as diadochokinesis, also are frequently used as essential criteria for the diagnosis of CAS (ASHA, 2007). The results of one study of school-aged children with CAS versus children with speech delays indicated that multisyllabic and non-word repetitions were significantly more impaired among children with CAS (Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004).

Prosodic characteristics have been studied as proposed diagnostic markers of CAS including: differences in rate, variability in pitch and loudness, as well as variations in speech-pause time (Shriberg, Aram, & Kwiatkowski, 1997b; Shriberg, Aram, & Kwiatkowski, 1997c; Shriberg, Campbell, Karlsson, Brown, McSweeny, & Nadler, 2003). These variations result in perceived atypical lexical and phrasal stress in speech produced by children with CAS (Shriberg et al., 2003a). These researchers documented excess and/or equal stress placed on both syllables of two-syllable words with stress placed on the initial syllable (i.e., trochaic words) (Shriberg et

al., 2003a). Munson, Bjorum, and Windsor (2003) found similar perceptual results on non-words produced by children with CAS. However, when acoustic analysis was performed, stressed syllables were not quantitatively different from their original standards. After reviewing the data, the researchers reported that the perceptual results might be linked more to frequency of the vowels rather than amplitude (Munson, Bjorum, & Windsor, 2003). Research also has been conducted to document differences between CAS and adult acquired apraxia of speech in regards to lexical stress (Odell & Shriberg, 2001). Perceptual findings indicated that prosodic features of stress remained excessively equal among children with CAS. However, the data showed inappropriate phrasing and rate among adults with acquired apraxia of speech that was not observed in children with CAS.

Variations in speech-pause time have been explored in regards to children diagnosed with CAS (Shriberg, Green, Campbell, McSweeny, & Scheer, 2003b). Findings suggested that speech-pause time was more variable among participants who were diagnosed with CAS; however, results were inconsistent. This variability typically was caused by a reduction in the duration of speech events and an extension of pause time in the utterances produced (Shriberg, et al., 2003b).

Clinicians have been known to hold differing opinions regarding diagnostic characteristics of CAS. In one study, clinicians were asked to indicate at least 3 characteristics that they believed contributed to the CAS diagnosis (ASHA, 2007; Forrest, 2003). The six most frequently cited criteria were inconsistent speech productions, general oral-motor difficulties, groping, inability to imitate sounds, increased errors corresponding to utterance length, and poor sequencing of sounds (Forrest, 2003). There were, however, a large number of responses described by the participants that included both associated and disproven diagnostic

characteristics. Although the current definition of CAS is not conclusive, the definition mainly focuses on variability in speech production including (but not limited to) variability in production of consonants and vowels and sequences of sounds (ASHA, 2007).

Summary and Rationale

Information on consistency of lip and jaw movement and acoustics during productions of repeated utterances for children with CAS is not available. However, research to date has identified perceptual speech abnormalities including speech-pause time and lexical stress as potential diagnostic markers for CAS. This research will further investigate the comparison of each of these characteristics in a child with CAS versus a TD child in an effort to quantify any or all differences exhibited.

The goal of this research is to determine whether kinematic and/or acoustic characteristics can potentially differentiate children with CAS from TD children without speech sound disorders. These characteristics must meet appropriate levels of sensitivity and specificity to be appropriate for clinical intervention and research. The benefit of the current research is to provide information regarding the effectiveness of study measures to identify differences in motor speech characteristics of children diagnosed with CAS according to the current ASHA (2007) guidelines versus TD children. More precise information regarding motor-speech planning skills in CAS and the resulting physical behaviors may provide greater insight into the process of the disorder and enhance knowledge regarding differential diagnosis. In addition, specific knowledge regarding motor skills may be useful in developing intervention procedures targeting CAS-specific abilities or deficits. The overarching goal of this project is to move toward a common speech protocol for use by investigators that in the future may also be translated for use in clinical interventions.

Plan of Study and Experimental Questions

For this project, kinematic and acoustic analyses provided insight into the stability of lip movement, prosody, and possible abnormalities in speech production in a participant with CAS and a TD participant. The following experimental questions were answered: (1) Are lip movements less consistent for the participant with CAS versus the TD participant; (2) Does the participant with CAS exhibit an increase in pause time percentages versus the TD participant; (3) Does the participant with CAS display abnormal stress as compared to TD participant in the repetition of trochaic words. The hypothesis is that the participant with CAS will produce greater variability in lip movements, exhibit greater percentages of pause time during speech than the TD participant, and demonstrate inconsistency and/or abnormality in lexical stress in trochaic word repetitions not seen in the participant with TD speech.

Chapter II: Method

Participants

Participants were selected from personal acquaintances and East Carolina University Speech-Language and Hearing Clinic clientele. The inclusion criteria for participation were as follows: (1) hearing within normal limits on audiometric screening; (2) corrected visual acuity sufficient to interpret stimuli presented in the study; (3) identified as TD or diagnosed with CAS, (4) able to read a short, simple passage (1^{st} grade level); (5) able to speak in a conversation for 5-10 minutes; (6) be 8 years or older; and (7) native speakers of American English, to reduce potential variability resulting from production of non-English words. Participants were excluded who presented with any of the following characteristics: (1) neuromuscular impairments (e.g., dysarthria); (2) TD but receiving special services in their educational program; (3) visual impairments that limit ability to read presented stimuli; and (4) reported cognitive impairments. There was no enrollment restriction based on gender, race, or ethnic origin.

During the initial meeting, consent and assent forms were obtained and then full eligibility was determined using the following assessments. A hearing screening was administered at 20dB HL for 1000Hz, 2000Hz, and 4000Hz in order to rule out a significant hearing loss that may influence a participant's performance. The *Oral Speech Mechanism Screening Examination* (St. Louis & Ruscello, 1981) was administered and a review of medical case history was used to rule out any craniofacial differences that may interfere with task performance and data collection. The *Peabody Picture Vocabulary Test, 4^{th} edition* (Dunn & Dunn, 2007) was administered to assess receptive vocabulary skills for all participants. The *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000) and the *Khan-Lewis Phonological Analysis-2* (Khan & Lewis, 2002) were administered twice to assess articulation

and phonological abilities, stability, and the phonetic inventory of the participants. The *Oral Speech and Sequencing Subtest* from the *Verbal Motor Production Assessment for Children* (Hayden & Square, 1999) was administered to rule out dysarthria. An assessment of all English vowels was administered to determine consistency of vowel production. In the vowel assessment, participants were asked to name pictures of common objects, eliciting all vowels and diphthongs in both open and closed syllables, in monosyllabic and disyllabic words.

Assessment results for both participants are presented in Table 1. The TD participant, an 8;0 year old female, presented with no deviations on the oral motor screening and passed a hearing screening. She exhibited age-appropriate picture naming on the *Peabody Picture Vocabulary Test, 4th edition* (Dunn & Dunn, 2007) with a standard score of 119. The participant demonstrated age-appropriate and consistent articulation based on the *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000) with no errors and no phonological processes present in the *Khan-Lewis Phonological Analysis-2* (Khan & Lewis, 2002). She presented with no signs or symptoms of dysarthria in the *Oral Speech and Sequencing Subtest* from the *Verbal Motor Production Assessment for Children* (Hayden & Square, 1999). No vowel errors were observed.

The second participant was previously diagnosed with CAS, which was reaffirmed using multiple levels of assessment. The participant with CAS, an 8;2 year old female, presented with no deviations on the oral motor screen other than noted rhythm deficits with repetitions of diadochokinetic strings. The participant also passed a hearing screening. She exhibited age-appropriate picture naming on the *Peabody Picture Vocabulary Test, 4th edition* (Dunn & Dunn, 2007) with a standard score of 109. She presented with inappropriate and inconsistent articulation errors based on the *Goldman-Fristoe Test of Articulation-2* (Goldman & Fristoe, 2000). Initially, the participant presented with five calculated errors, four vowel errors, and

abnormal perceptual stress on seven words. On the subsequent presentation, the participant produced three calculated errors, seven vowel errors, and abnormal perceptual stress on five words. No consistent phonological processes were noted in the *Khan-Lewis Phonological Analysis-2* (Khan & Lewis, 2002). She presented with no signs or symptoms of dysarthria in the *Oral Speech and Sequencing Subtest* from the *Verbal Motor Production Assessment for Children* (Hayden & Square, 1999). Two vowel errors were noted in the vowel assessment at the word level and six were noted at the sentence level. Perceptual syllabification errors at both the word and sentence levels were noted. A clinical committee consisting of two certified speech-language pathologists familiar with CAS verified the given diagnosis of CAS through observation of assessment and results. Both were in agreement with the stated diagnosis.

Per guidelines stated for participation, each of the above participants qualified, as they presented with all inclusionary and no exclusionary criterion. Participants were also chosen based on their proximity in age, since research to date has shown minimal within age group variations for speech motion consistency (Green, et al., 2002). The TD participant presented with no noted deviations on any tasks presented. The participant diagnosed with CAS met guidelines for participation with inappropriate and inconsistent articulatory abilities, vowel errors, and abnormal perceptual stress as evidenced by Table 1 below.

	TD Participant	**Participant with CAS**
Oral Speech Mechanism Screening Examination	No deviations	Deviations only on repetition of diadochokinetic strings
Standard Score: *Peabody Picture Vocabulary Test*	119	109
Errors on initial *Goldman-Fristoe Test of Articulation*	0	5 consonant 4 vowel
Errors on subsequent *Goldman-Fristoe Test of Articulation*	0	3 consonant 7 vowel
Phonological processes on the *Khan-Lewis Phonological Analysis*	0	0
Signs of dysarthria on the *Oral Speech and Sequencing Subtest* from the *Verbal Motor Production Assessment for Children*	None	None
Errors on Vowel Assessment: Word Level	0	2
Errors on Vowel Assessment: Sentence Level	0	6

Table 1: Participant Qualifications for Participation

Stimuli and Procedure

Data collected from each participant included sentence repetition, a brief story retell, and trochaic word repetition. During data collection, participants were seated upright in a reclining chair with tasks presented via a projected image (Epson PowerLite S5 Multimedia Projector) onto a projection screen (40”x55”) in front of them. Simultaneous digital recordings of 3D movement traces and digital audio and video recordings of all tasks were obtained for both participants. Overall, the participants read the sentence “Buy Bobby a puppy” eighteen times, although this task was divided into three segments of six repetitions and presented between other data collection. Specifically, participants read the item number and the sentence aloud (e.g., “One. Buy Bobby a puppy; Two. Buy Bobby a puppy.”). Data from this task were used for calculation of the STI. The story *Bats, Beets, and Boots* (Speech Production Lab & Jackson,

2004) was presented with images while the story was presented by the examiner. Participants subsequently retold the story in their own words. These data allowed for calculation of speech-pause time. The participants repeated the following trochaic words after the examiner's model in order to assess lexical stress: "chicken, dishes, hammer, ladder, peanut, puppy, robot, and window" (Shriberg et al., 2003a). Data collection sessions typically lasted approximately 30 minutes in entirety, with only one session per participant.

Recording Channels

Data were collected in a motion analysis laboratory equipped for audio, video, and kinematic recording. Participants' acoustic and visual information was obtained through digital recordings (Marantz PMD660 audio recorder) and a digital video recorder (Sony Handycam High Definition Digital Video Recorder). The audio component of each digital video file was recorded at 44.1 kHz (16-bit) using a miniature professional-quality earset microphone (Countryman Isomax E6) that was placed one inch away from the mandible. In this manner, mouth-to-microphone distance was controlled for each participant. Lip and jaw movements were recorded using a 3D high resolution, optical motion capture system (Qualisys Oqus Motion Analysis System). This system captured the position of reflective markers placed on facial structures using four digital cameras. The system software computed three-dimensional positions based on the two-dimensional views provided by each of the four cameras.

Eleven reflective markers, 3 mm in diameter, were attached with adhesive to each participant's facial structures producing targeted movements as shown in Figure 1. Reference points were placed on the forehead using a non-symmetrical array of 4 markers placed in a cross-like fashion embedded in an off-the-head array. For this study, data were obtained from the

markers on the lips (4 markers), the chin (1 marker), and the jaw (2 markers). The markers in the off-the-head array were used solely as reference points.

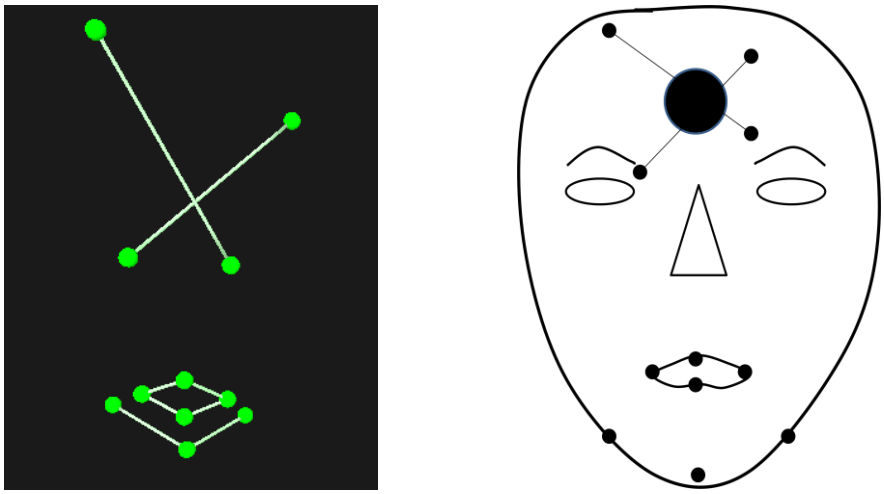

Figure 1: 2D Display of Facial Structures with Schematic Placement of Reflective Markers

Data Analysis

Kinematic data were reduced and analyzed using customized algorithms developed for Matlab software (Matlab [Computer Software], 2004). The kinematic analysis relied on computer-assisted routines and was completed using the Speech Movement Analysis and Spatial Histograms (SMASH) program (Green et al., 2002). This program was designed to improve measurement reliability and to maximize the information that can be extracted from the large data set generated by each participant. The program allows for consideration of head movement by subtracting the analyzed markers (i.e., lips) from the stability markers in the off-the-head array placed at the forehead. Prior to entering the data into the software, each of the eighteen repetitions of "Buy Bobby a puppy" was saved separately. The start time of the sentence was set as the frame prior to the initial negative movement of the lower lip on the /b/ of "buy." The end

time of each sentence was set as the frame prior to the negative movement of the lower lip on the second /p/ of the word “puppy.” This allowed for maximum consistency in the subsequent calculation of STI.

From the reduced data, the STI was factored on the repeated sentence “Buy Bobby a puppy.” Initially, the sentence data were overlapped in order to observe the consistency and stability of the signals. SMASH allowed for computerized amplitude normalization by dividing each movement trace by its standard deviation (Smith & Goffman, 1998). Linear temporal normalization was achieved via the same program with interpolation of each signal onto a time base of 1000 points using computerized algorithms (Green et al., 2002). Lastly, the sum of standard deviation of lip movement at a set number of intervals (2%) across the normalized signal was used to calculate the final STI (Smith et al., 1995). These calculations were directly computed using the SMASH program (Green, Moore, & Reilly, 2002). The calculations of all eighteen repetitions were analyzed as a whole. According to current research; fatigue is not a substantial variable for orofacial musculature involved in speech and/or non-speech movements (Solomon, 2006); however, to minimize any risk of potential fatigue, the participants completed the task in 3 blocks of 6 productions each.

Acoustic analyses were performed specifically related to speech-pause time and lexical stress. The speech-pause time in story retell was calculated by comparing ratios of total speech time versus total pause time. Adobe Audition (Adobe Systems, Inc., 2010) was used to compile the data. The files were uploaded into the software program and visual and perceptual measures were used to determine speech time, removing time for pauses both between and within words. After total speech time was determined, the total length of each utterance was measured. These measurements were summed to create a total length of all utterances. The speech time was then

subtracted from the total length of all utterances to yield total pause time. From these data, percentages of speech time versus pause time were calculated for each participant.

Frequency, amplitude, and duration of the vowel were used to examine the lexical stress placed on the trochaic word syllables by each participant. When calculating the duration of each vowel, similar procedures were followed as that of Shriberg et al. (2003a); therefore, factoring vowel length included the nasalized portions of the vowel. The average frequency and amplitude of the vowel was calculated using Praat (Boersma & Weenink, 2009), an acoustic analysis computer program. Each of these measures was used to form a ratio of the first syllable over the second syllable of each word (Shriberg et al., 2003a). Each aspect of lexical stress including frequency, amplitude, and vowel duration was examined.

Chapter III: Results

Descriptive statistics were used to analyze data for both participants, as this was exploratory research to determine whether the current methods could be used to effectively distinguish and diagnose CAS. The following analyses involved direct comparisons between participants on the STI, speech-pause time, and lexical stress variables.

Spatiotemporal Index

Each participant's eighteen captured repetitions were used in the calculation of STI (i.e., sum of standard deviations of the eighteen signals at 2% intervals) to evaluate kinematic data of lip motion consistency during speech. Thus, all eighteen repetitions were used in the calculation of STI. These data were used to investigate and compare consistency of lip movements for the two participants. There was a trend towards less consistency in movements for the participant with CAS than the TD participant. For the TD participant, the calculated STI for the lower lip was 36.705. The upper portion of Figure 2 illustrates all eighteen traces of lower lip movement for the TD participant during production of the target sentence in displacement across the 1000-point interpolated time base. The standard deviations at 2% intervals used in the calculation of the STI of the TD participant's lower lip are illustrated in the lower portion of Figure 2.

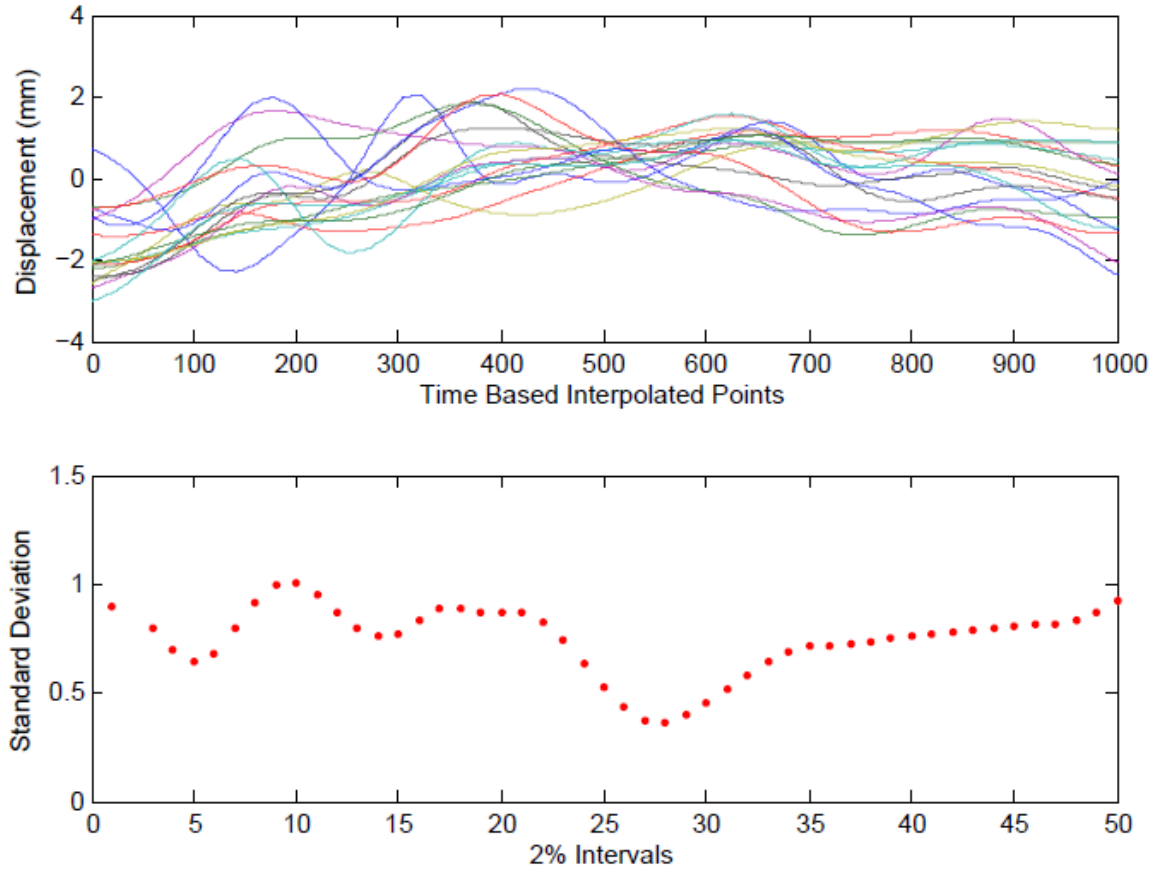

Figure 2: TD Participant's Lower Lip Movement Tracings and STI Intervals

The calculated STI for the TD participant's upper lip was 36.712. While the upper portion of Figure 3 illustrates all eighteen movement traces of the upper lip for the TD participant for each production of the target sentence, the lower portion charts the standard deviation at 2% intervals used in the calculation of the STI for the identical data.

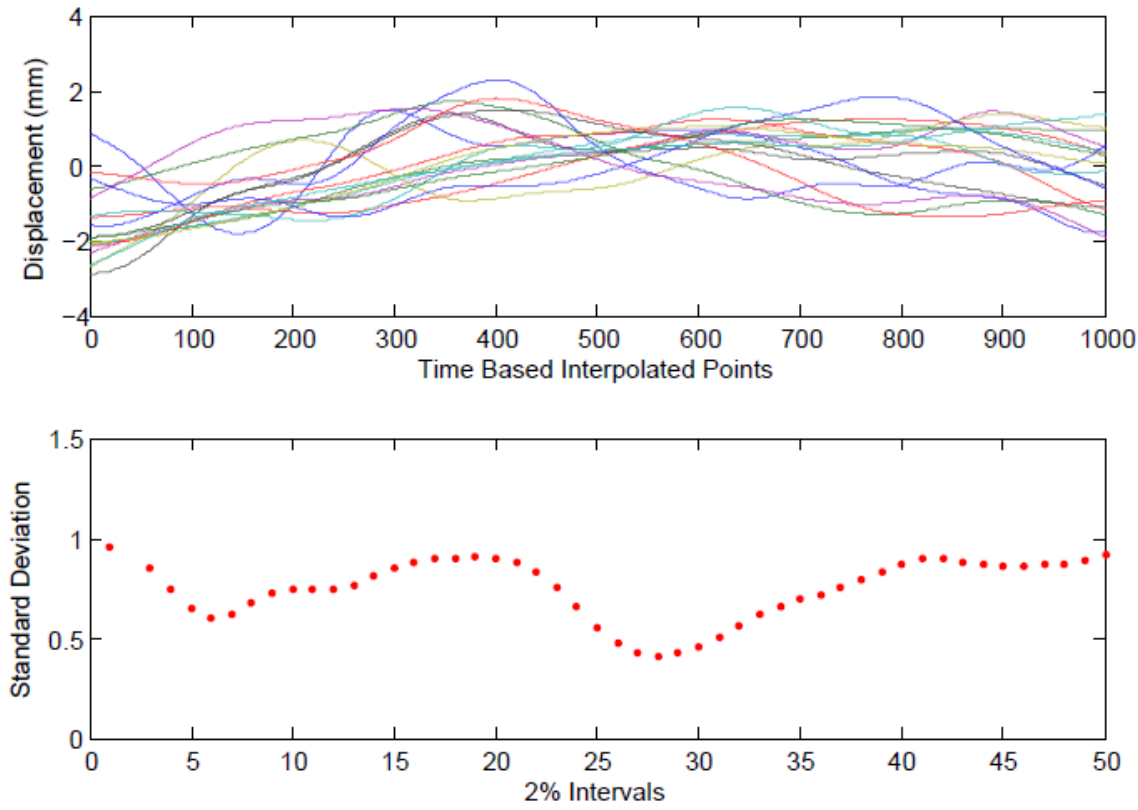

Figure 3: TD Participant's Upper Lip Movement Tracings and STI Intervals

For the participant diagnosed with CAS, the STI value for the lower lip was calculated as 44.035. Figure 4 illustrates the movement tracings of the participant with CAS' lower lip during each of the eighteen repetitions of the target sentence. The lower portion of Figure 4 plots the standard deviation at 2% intervals used in the calculation of the STI derived from movement of the lower lip of the participant with CAS.

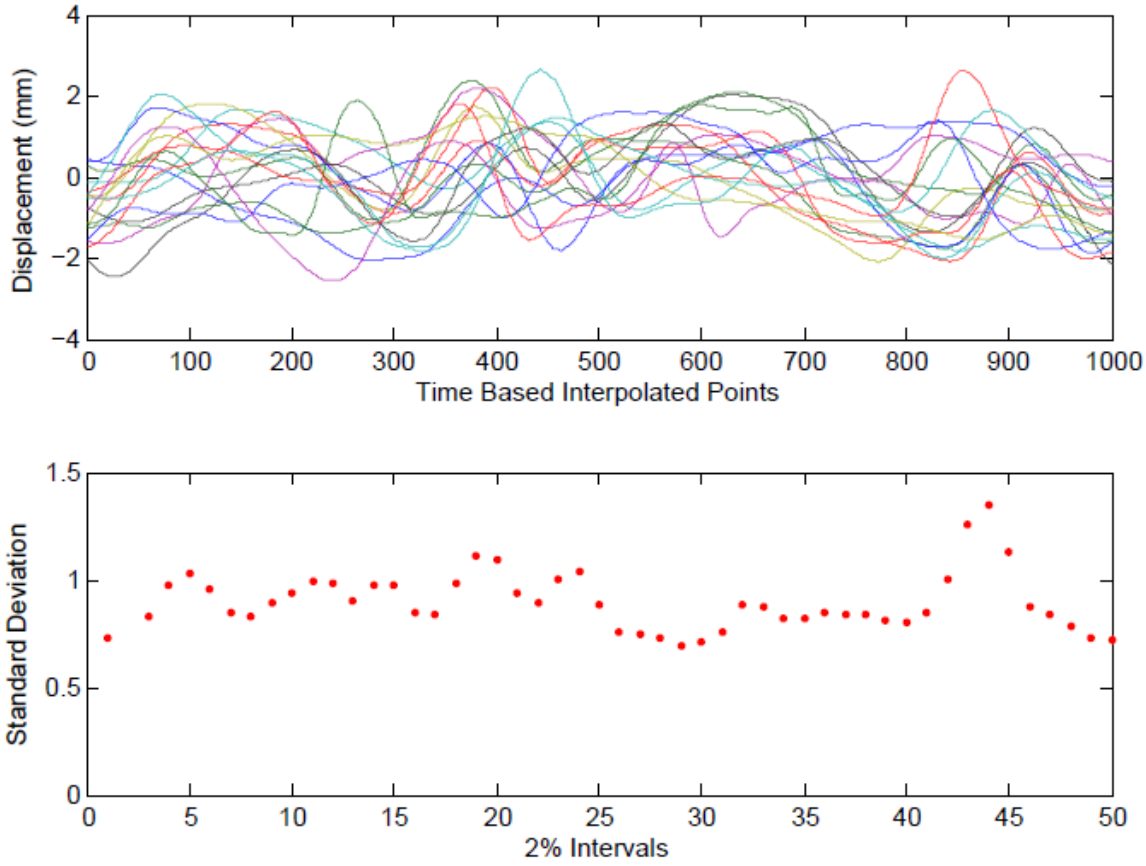

Figure 4: CAS Participant's Lower Lip Movement and STI Intervals

The STI for the upper lip of the participant with CAS during the repetitions was calculated as 45.559. The upper portion of Figure 5 displays the movement tracings of the participant with CAS's upper lip while the lower portion plots the standard deviation at 2% intervals used in the calculation of the STI of the same data.

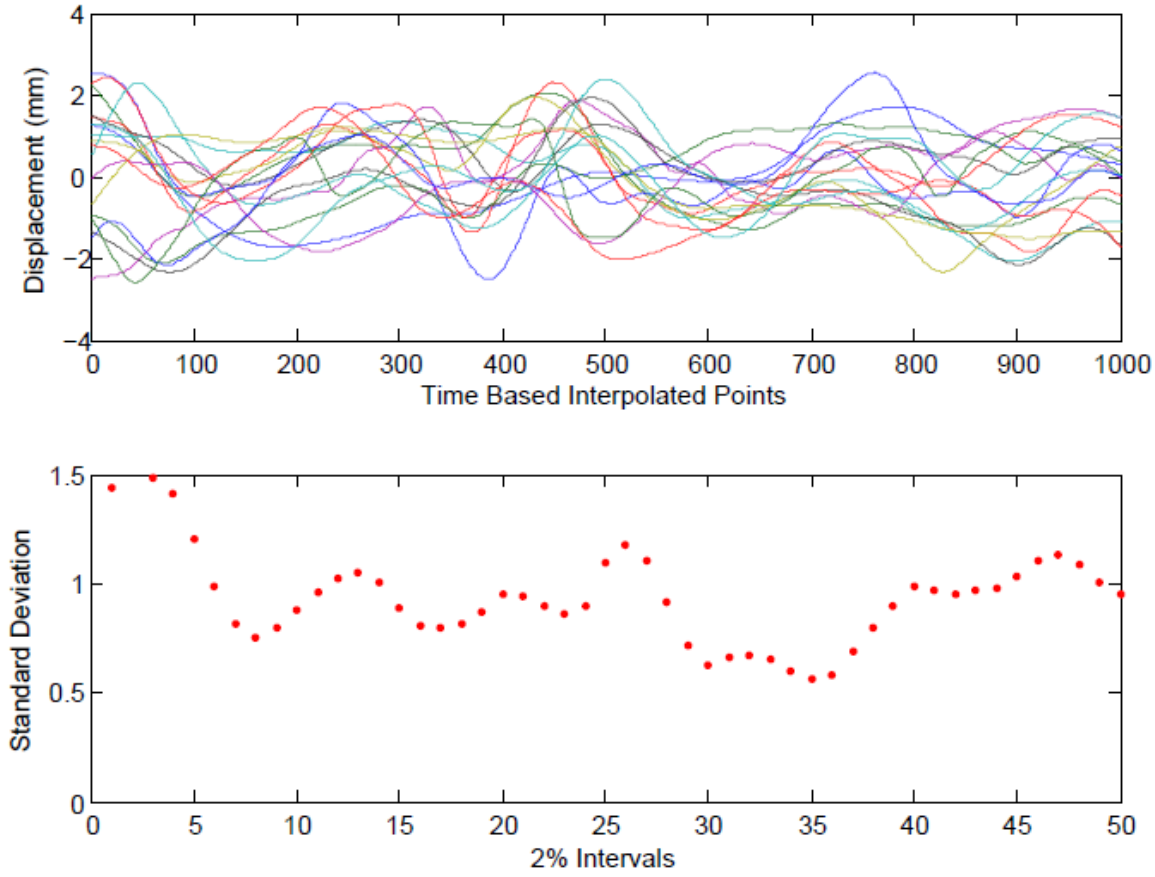

Figure 5: CAS Participant's Upper Lip Movement Tracings and STI Intervals

As stated previously, a lower calculated STI indicated lower variability on speech movements produced during production of the sentence "Buy Bobby a puppy." These data show a 7.330-point increase in lower lip variability for the participant with CAS over the TD participant. However, the data also reveal a similar, but slightly larger (8.848-point) difference in upper lip movement variability for the participant with CAS over the TD participant.

Speech-Pause Time

Story retelling data were used to examine whether the participant with CAS displayed larger percentages of pause versus speech time than the TD participant. Results indicated that cumulative speech and pause times were longer for the participant with CAS, whose total retell

time (excluding between-image/sentence pauses) was 37.29 seconds, with 13.275 seconds of pause time. The TD participant retold the given story in 17.192 seconds with only 4.495 seconds of pause time. Figure 6 illustrates the total lengths of the story retell divided into speech and pause time for each participant.

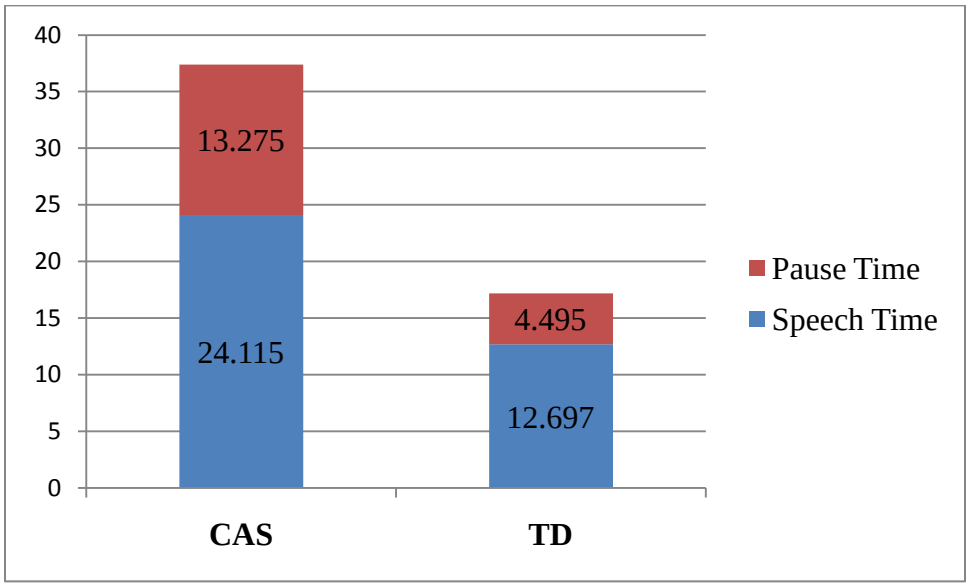

Figure 6: Total Length of Utterance per Participant with Speech and Pause Times Noted

When overall speech-pause time was compared within each participant's production, the participant with CAS had 35% pause time while the TD participant exhibited 26% pause time. Figure 7 charts the percentages of speech versus pause time for each participant.

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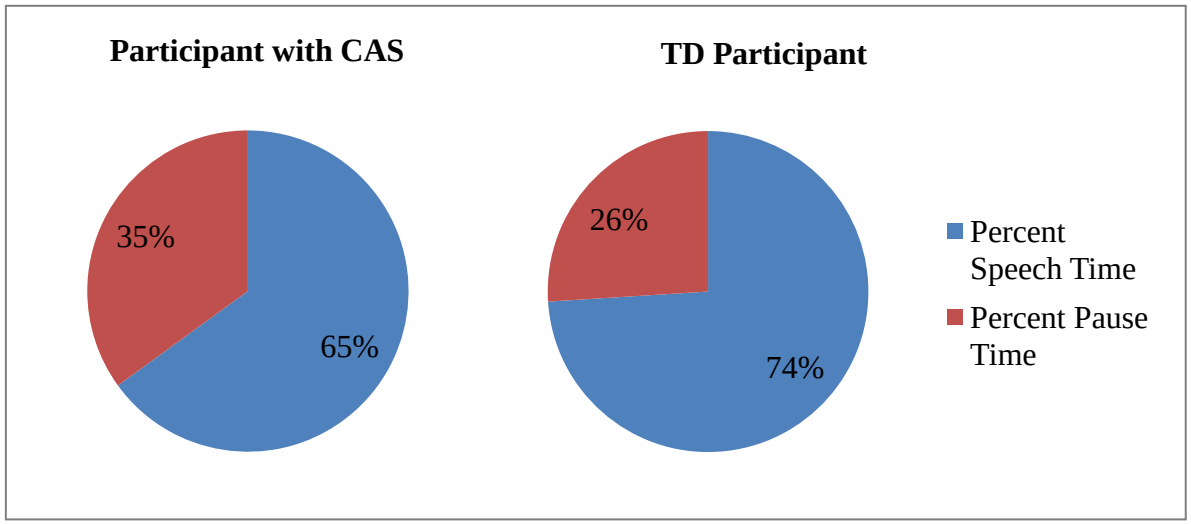

Figure 7: Percentage of Speech versus Pause per Participant

Lexical Stress

Acoustic signals recorded during the repetition of eight trochaic words were used to determine whether the participant with CAS exhibited differences in lexical stress when compared with the TD participant. Sixteen vowels were analyzed for each participant; however, one of the second-syllable productions for the TD participant was not analyzed for frequency because the formants were not well established. For the ratio of vowel length of the first syllable over the second, the TD participant presented with a ratio of 1.442, whereas the participant with CAS presented with a ratio of 0.769. Figure 8 illustrates vowel length ratios calculated for each trochaic word.

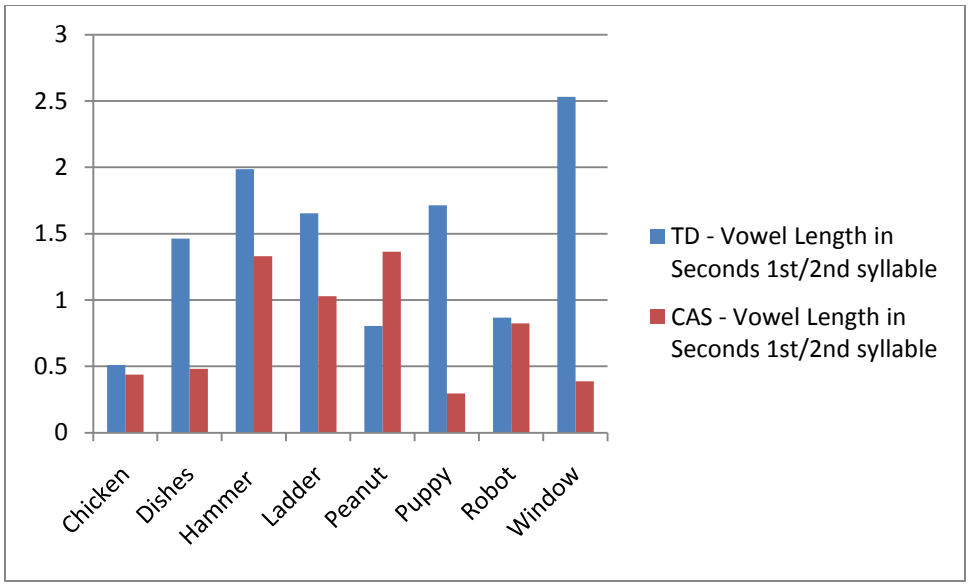

Figure 8: Vowel Length in Seconds per Trochaic Word: TD Participant versus Participant with CAS

The mean frequency ratio for the TD participant was 1.216 versus 0.986 for the participant with CAS. Results for each word individually were similar excluding the word “dishes” in which the TD participant exhibited a much higher frequency ratio (2.07) as compared to the participant with CAS (1.16). The word “puppy” for the TD participant was not analyzed for frequency, since the formants were not sufficiently established. This occurrence was most likely due to the nature of the word ending with a voiceless plosive. Figure 9 illustrates the ratio of frequency of the first syllable over the second syllable for each trochaic word.

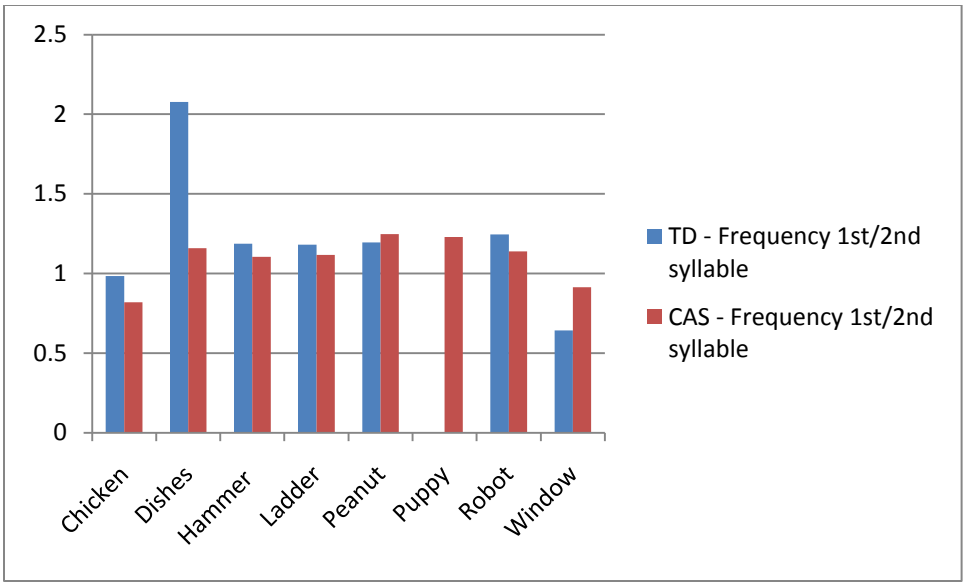

Figure 9: Frequency per Trochaic Word: TD Participant versus Participant with CAS

Similar ratios were found for mean amplitude with 1.175 and 1.131 respectively. No differences were noted between the two subjects in regards to amplitude. Figure 10 illustrates the ratios for amplitude per trochaic word.

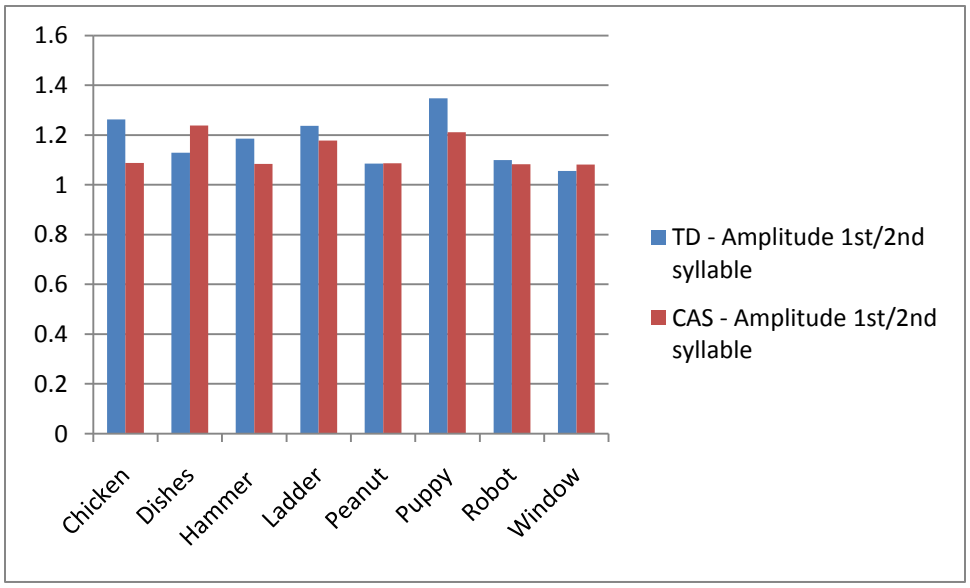

Figure 10: Amplitude per Trochaic Word: TD Participant versus Participant with CAS

Neither frequency nor amplitude ratios indicated observed differences between the two participants. The key quantitative differences in lexical stress were found in regards to vowel length. These vowel length data indicate that, on average, the participant with CAS exhibited shorter vowel duration on the initial syllable versus the second. This signifies atypical lexical stress, since on trochaic words initial vowels typically are lengthened to add stress to the initial syllable.

Intra-judge reliability was calculated by re-computing vowel length, frequency, and amplitude on 12.5% of the trochaic words, which included one initial and one final vowel for each participant. The mean difference noted was 0.116. This was considered to indicate sufficient reliability given the somewhat subjective nature of determination of the beginning and ending of the vowel produced.

Chapter IV: Discussion

The current research investigated methods for potential feasibility in differentiating speech characteristics of TD children from those with CAS. The methods used included kinematic and acoustic analyses to evaluate speech motion stability by calculating the STI in repetitions of a target sentence, speech versus pause timing in story retell, and lexical stress in trochaic word repetitions. This research identified differences between the two participants in all three areas. It appears that the procedures used may be an effective, objective method for differentiating children with CAS from TD children.

Spatiotemporal Index

Results of this research indicate that the participant with CAS exhibited a larger calculated STI on sentence repetition (e.g., "Buy Bobby a puppy"). These data indicate that upper and lower lip movements were less consistent for the participant with CAS versus the TD participant after analyzing the overlapped repetitions of the target sentence. Although no research to date has examined the kinematic stability or calculated STI for children with CAS, this variable has been completed for TD children. In data recording, Sadagopan and Smith (2008) observed that the STI of a TD 5-year-old during the repetition of "Buy Bobby a puppy" in isolation was 17.36. However, when the sentence was embedded in a more complex sentence, the STI rose to 33.59 and 35.72 depending on complexity. The STI values in the current research correspond more closely to Sadagopan and Smith's (2008) data calculated during the repetition of the target sentence in a complex utterance than production in isolation. This may be due to the inclusion of the item number during production of each target sentence, thus introducing greater complexity of task production since each number is different and thus results in a different motor plan for production, rather than simply repeating the same sentence in isolation. Additionally, the

data comparisons reported in this research used identical procedures for computing STI from kinematic data and are thus considered an accurate reflection of both participants' performance.

Review of the literature revealed no data in regards to kinematic analysis of upper lip movements and calculation of the corresponding STI for TD children or children with CAS, thus limiting broader comparisons. The results of this research found minimal differences within participants in regards to the upper and lower lip movements during repetitions of the target sentence. The extent of differences identified between participants' upper lip STIs were similar to the difference found between the lower lip STIs. These data indicate that movements of the upper lip were related to those of the lower lip for both participants. This strengthens the argument that CAS is a speech motor production disorder that involves the entire speech production system and indicates that upper lip movements may be another potential measurement site for future research.

Speech-Pause Time

Data analyzed from participants' story retell identified that the total length of all combined utterances was greater for the participant with CAS. The reason for this extended total speech time is unclear; however, task variability might have been an influencing factor. Further analysis of the participants' story retell indicated greater pause time within and between words for the participant with CAS. This adds credence to the argument for a motor programming impairment associated with CAS. With a motor programming impairment, a child might require increased time to form and sequence sounds to produce words and sentences. Variations in stress and prosody, often clinically reported with CAS, may either increase pause time during speech production or in fact, be directly caused by longer pause times. Although no numeric data were accessible for direct comparison of speech versus pause times for either the TD participant or the

participant with CAS, these data are consistent with previous research. Using another measurement technique known as the coefficient of variation, Shriberg, et al. (2003b) found an increase in pause time among children with CAS versus TD children.

Lexical Stress

As expected, lexical stress data showed relatively similar mean frequency and amplitude ratios between the initial and second syllable of trochaic words. However, vowel data indicated a lengthier initial syllable production than that of the second syllable for the participant with CAS. These vowel length data indicate that on average, the participant with CAS exhibited greater lexical stress in regards to vowel length on the second syllable. This does not correspond to typical trochaic word lexical stress productions and may be indicative of a unique pattern associated with CAS. These findings, again, may be related to a motor programming aspect of the CAS disorder, specifically related to the ability of the child to move articulators in such a way to form the sounds accurately, rapidly, and consistently. These data further support the theory that CAS is a movement disorder, since vowel length rather than amplitude or frequency abnormalities were found. Vowel length may be the key perceptual difference that clinical speech-language pathologists have reported to distinguish the speech of CAS from TD children (McCabe, et al., 1998). The findings of the current research are congruent with research conducted by Shriberg, et al. (2003a) and Munson, Bjorum, and Windsor (2003) who indicated a disturbance in lexical stress for children with CAS. However, Munson, Bjorum, and Windsor (2003) reported that perceptual changes may be related to frequency abnormalities which were not shown in the given research.

Limitations

An obvious limitation in the current research was the size of the study. Having only one participant in each category made the data obtained impossible to generalize to a larger population. This limitation was considered from the onset; however, it was concluded that this research would remain exploratory to investigate the feasibility of the combined measures for examining speech of children with CAS.

Another foreseeable limitation in the current research continues to be the questioned diagnostic characteristics of CAS and thus, those used to qualify the participant with CAS to participate in the study. Research has led to CAS being diagnosed mainly by features such as inconsistency in speech productions over time, as well as vowel errors and perceptual speech differences (ASHA, 2007). These same variables were used as the main qualifying factors after ruling out dysarthria, physical (i.e., craniofacial), or phonological speech sound disorders. Critical to the use of the measures in this study was avoidance of the circular argument often identified in previous studies, in which researchers attempted to measure the very features used as inclusion criteria. One key factor in selection of the independent variables was that they be measures not specifically used as inclusion criteria but instead targeted quantitative measurement of these clinical/perceptual features or components of speech suspected as influences (e.g., inconsistent speech production inclusion criteria were obtained from speech samples while STI was used to calculate consistency of lip movements during speech).

A possible weakness in the current research relates to the specific trochaic words selected for the study. Although previously used to compute lexical stress, the tasks were found to be difficult to analyze, likely due to co-articulatory interference from nasalized vowels and diphthongs. This may have interfered with data interpretation. Selecting trochaic words without

these features in future research may allow for easier and more accurate acoustic vowel calculations. Although this is a limitation, the impact was equal for both participants in the current study.

Implications for Future Research

Additional larger scale studies are needed to confirm the findings of the current research and to create standardized measurements of reference for STI, speech-pause time, and lexical stress ratios. Further research may explore the impact of age on the given factors in regards to children with CAS versus TD children, both as moments in time and longitudinally. This might provide insight into the similarities and differences observed in STI with age compared to those which have already been documented for TD children. Such knowledge may provide a greater understanding into the timing of diagnosis of CAS. Further exploration of STI when producing other sentences and in conversation also may be beneficial in showing how STI differences occur in children with CAS and allow for a broader view of what factors influence differences in STI. By further exploring other sentences, a putative feature may be found that is more definitive in the diagnosis of CAS. Analysis of the video signal without the acoustic signal present might reveal valuable data about whether motion or stability characteristics could be identified visually without the use of kinematic or acoustic analyses.

Another option for research might include analysis of speech-pause time and lexical stress on the same data used for kinematic analysis (e.g., “Buy Bobby a puppy”). These data may identify whether stability in speech movements impacts speech-pause time and lexical stress, thus simultaneously combined to evaluate relative contributions of each to the overall characterization of CAS. The use of STI as an evaluation of the effectiveness of motor learning therapies for children with CAS has yet to be examined. These data may assist in determining

which techniques and therapies are best suited to address the motor production issues seen in children with CAS.

Examination of the influence of spontaneous speech versus scripted or read responses may provide additional information on speech-pause timing. This may add information regarding whether speech production differences also occur during reading tasks and if present, what types of influence the reading task has on qualitative and quantitative measures of speech characteristics in children with CAS.

When examining lexical stress, this research documented the feasibility of identifying inconsistencies of speech productions among children with CAS when repeating the same word. To fully understand the impact of lexical stress as a possible diagnostic feature of CAS beyond single word repetition, it is necessary to research lexical stress analyses in conversational and spontaneous speech production.

Additional studies are needed which include examination of performance on kinematic and acoustic tasks applied to other speech sound disorders (e.g., phonological process, articulation disorders) that are potentially misdiagnosed as CAS. Research of this nature may add data to differentiate not only between speech characteristics of CAS and TD children, but begin to elucidate specific features distinguishing among other speech sound production disorders.

Summary and Conclusions

In the current investigation, the participant with CAS presented with greater variability of lip movements during repetitions of the target sentence, as exhibited by a larger factored STI. Analysis of the acoustic signal during production of a story retell task revealed that the participant with CAS exhibited lengthier overall story retell time as well as longer pause versus speech time than the TD participant. Lastly, during repetition of eight trochaic words, the

participant with CAS presented atypical mean lexical stress associated with greater vowel length on the second syllable; the TD participant displayed a more typical pattern, exhibiting stress primarily on the initial syllable. In sum, the data collected in this research indicate feasibility for implementing STI, speech-pause time, and lexical stress measures to differentiate TD children from children with CAS in kinematic analysis.

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Appendix: Approval

UNIVERSITY AND MEDICAL CENTER INSTITUTIONAL REVIEW BOARD
HUMAN BEHAVIORAL AND SOCIAL SCIENCE INTERNAL PROCESSING FORM
SUBMISSION FOR UMCIRB REVIEW
FULL AND EXPEDITED RESEARCH

DEMOGRAPHIC INFORMATION

Type of application : ☑ **New** ☐ **Modification Date : June 2009 UMCIRB # :** 09-0234

Title of proposed research (this title must match protocol, funding application and consent form) : Speech Production Analyses : Characterizing Typical Development versus Childhood Apraxia of Speech

Principal Investigator, credentials, department, section and school :
Laura J Ball CCC-SLP; Associate Professor, Communication Sciences and Disorders, School of Allied Health
Check the institutions for which the principal investigator is associated : ☑**ECU** ☐**PCMH** ☐**Other**

Subinvestigators, credentials, department, section and schools :
Jennifer M. Lemkes, Graduate Student, Communication Sciences and Disorders, CSDI, School of Allied Health

*** **Investigators not associated with ECU or PCMH require submission of an Unaffiliated Investigator Agreement.**

List of all items related to this research study submitted for UMCIRB review and approval : Internal processing form, consent (1 legal representative), assent (1 child)

SOURCE OF FUNDING

☐ **Government Agency, Name** :
☐ **Private Agency, Name** :
☐ **Institution or Department Sponsor, Name :**
☑ **No funding**
☐ **Grant : include 3 copies of the final grant application for full committee reviews or 1 copy for expedited reviews**

Fund number for IRB fee collection (applies to all for-profit, private industry or pharmaceutical company sponsored projects) :

Fund	Organization	Account	Program	Activity (optional)

		73059		

NOTE : The UMCIRB Conflict of Interest Disclosure Form needs to be submitted for expedited and full review.

CHECK ALL INSTITUTIONS OR SITES WHERE THIS RESEARCH STUDY WILL BE CONDUCTED :

☑ **East Carolina University**
☐ **Other**

CHECK ALL OF THE FOLLOWING INVOLVED IN THIS STUDY

Population Specifically Targeted

☑ **Normal volunteers**
☐ **Adults (> 18 yrs old)**
☑ **Minors (< 18 yrs old)**
☐ **Institutionalized Participants**
☐ **Students**
☐ **Participants at other Institutions**
☐ **Mentally Impaired Participants**
☐ **Prisoners**
Photography
☐ **Wards of the State**
☐ **Minorities**
☐ **Participant pools**
☐ **Staff Participants**
☐ **Non-English speaking**
☐ **Public School System**
☐ **Day care facilities**

Methods/Procedures

☑ **Surveys / Questionnaires**
☑ **Interviews**
☑ **Standardized Tests**
☑ **Non-standardized Tests**
☐ **Focus Groups**
☐ **Deception**
☐ **Databank Information**
☑ **Videotaping / Voice Recording /**

☐ **Public observation**

RESEACH RISK AND LEVEL OF REVIEW REQUIRED

Research participants will be placed at as defined below :
☑ **No more than minimal risk**
☐ **More than minimal risk**

Minimal risk means that the probability and magnitude of harm or discomfort anticipated in the research are not greater in and of themselves than those ordinarily encountered in daily life or during the performance of routine physical or psychological examinations or tests. 45 CFR 46.102(i) The definition for prisoners differs and is located at 45 CFR 46

What level of review does your proposal require?
☑ **Expedited**
☐ **Full**

Those research studies utilizing Pitt County Memorial Hospital resources, Brody School of Medicine resources or involving ionizing radiation should complete the Institutional Approval for Research Form**.**

Research Questions

1. Subject Selection

- **Describe how participants will be selected or recruited for the research, including enrollment procedure.** The participants will be selected from the ECU Speech, Language, and Hearing Clinic and other professional and/or personal acquaintances.

- **Identify the projected number of participants to be enrolled.** The projected number currently stands at 2; including 1 typically developing (TD) child and 1 with Childhood Apraxia of Speech (CAS).

- **Outline the inclusion and exclusion criteria for this research study**. The inclusion criteria for participation for all participants are : (1) hearing within normal limits on audiometric screening; (2) corrected visual acuity sufficient to interact with stimuli presented in the study; (5) identified as typically developing or diagnosed with CAS, (6) able to read a short, simple passage (2^{nd} grade level); (7) able to speak in conversation for 5-10 minutes; (8) be 8 years or older; and (9) native speakers of American English, to reduce potential variability resulting from production of non-English words. Potential participants will be excluded with : (1) neuromuscular impairments (e.g., dysarthria); (2) typically developing but receiving special services in their educational program; (3) visual impairments that limit ability to read presented stimuli; (4) reported cognitive impairments. There is no enrollment restriction based on gender, race, or ethnic origin.

- **Provide a justification for the sample size selected.** The projected number of participants was chosen because it is a proof of concept project to determine whether an 8-year-old child with CAS or who is TD will be distinguished using the strategies attempted.

- **Describe the safeguards in place to protect the rights and welfare of any vulnerable participants enrolled in this research study.** Because participants will be minors and therefore considered vulnerable, each participant's legal representative (e.g., parent, guardian) will be informed of all procedures and sign their consent for the minor child to participate. Additionally, child participants will be informed of all procedures in simple language and asked to indicate their own assent (either witnessed verbally or signed) to participate.

Are there any advertisements (public display in written, radio, or TV form) for participant recruitment?

☐ **Yes** ☑ **No If yes, attach the advertisements to the processing form.**

Does the research include any monetary inducements, compensation or reimbursement for participation in this research study?

☐ **Yes** ☑ **No If yes, attach the payment schedule to the processing form or provide specific protocol reference.**

Will the sponsor reimburse for any items or procedures or supply any items at no cost involved in this research study?

☐ **Yes** ☑ **No If yes, attach written documentation of the items that will be reimbursed or supplied by the sponsor unless this information is specifically noted in the research protocol.**

Are there any associated costs that participants will incur in as a result of participating in this research study?

☐ **Yes** ☑ **No If yes, describe these costs.**

2. Researcher Qualifications

- **Name and list the duties of the research team members and describe the qualifications of each member to perform their duties.** Both Dr. Laura J. Ball and Jennifer Lemkes will participate in the data collection. The study procedures include (1) placement of small reflective markers on the face and an over-the-ear microphone; (2) recording acoustic, kinematic, and aerodynamic measures during speech production; and (3) analysis of the data obtained (e.g., motion analysis, acoustic waveforms, phonatory transcription). Dr. Ball has completed research using the identical strategies in previous studies and prior to completion of the study; Ms. Lemkes will be fully trained to operate all of the equipment for both capture and analysis of the data.

- **Include the completion date of the human protections modules located on the UMCIRB web site.**
  Jennifer M. Lemkes – completed 9-19-08
  Laura J. Ball – completed 7-23-07

3. Risk Determination

- **Describe the research setting, listing any safeguards in place for participant safety.** The researchers will employ universal precautions throughout all data collection sessions. During data collection, participants will be seated upright in a comfortable recliner-type chair. 1-mm reflective markers (spheres) will be affixed to the face using double-sided tape (designed to for this purpose) that has low allergenic/irritancy impact. Skin condition will be monitored during the session for any irritation that may occur and removed immediately upon any observed effects. A small microphone will be placed over the ear, with the microphone placement approximately 5mm from the corner of the lips. A single-use, disposable nasal mask will be used for collection of aerodynamic measures through placement over the nose intermittently during select speech tasks. All stimuli will be presented either using verbal instructions and/or via a projected image (e.g., words or sentences to read, objects to identify) on a screen in front of them. All data will be

collected in the ECU motor-speech laboratory using established equipment that is calibrated. Simultaneous digital recording of 3D movement traces and digital video recordings of all tasks will be obtained for all participants. Orofacial movements will be recorded during spontaneous and purposeful, modeled utterances (i.e. facial gestures, vocalizations, and speech). Data collection sessions will typically last 30 minutes in entirety.

4. Risk Determination

- **Describe all foreseeable physical, psychological, economic, social, legal and dignitary risks to the participants, with steps outlined to minimize those risks. Risks should be described in terms of probability or likelihood, magnitude and duration when possible.** Investigators will take all measures possible to minimize any unforeseen risks that arise. Investigators will use standard, calibrated equipment that has been established in the field with no associated risk. Measures will also be taken in order to ensure that all persons involved on the research team will be fully trained to use the equipment.

- **Outline the mechanism for reporting adverse events or unanticipated risks to participants or others for this study.** If an adverse event were to occur, investigators will follow regulatory steps in reporting it to the proper authorities immediately.

5. Data/Safety Monitoring : Data monitoring includes activities such as interim analysis or other opportunities for both individual and aggregate study data to be reviewed to ensure the safety of participants. A plan for this type of data monitoring may be required to meet the criteria for IRB approval in order to ensure the protection of participants involved in the research, to review the risk-benefit analysis, and to ensure there are no new findings for which current or future participants should be apprised.

- **If applicable, describe how data will be reviewed to determine if the study procedures should be changed during the course of the study.** N/A

6. Anticipated Benefits

- **Describe the benefits of the research study to participants or others.** The benefit of this research study is to provide information regarding the ability of the study measures to identify differences in motor speech characteristics of children with CAS versus TD children. The investigation into exact motor-speech planning skills in CAS and the resulting physical behaviors may provide greater insight into the disease process and allow for greater knowledge regarding beneficial differential diagnosis and intervention procedures. This information is currently unavailable.

7. Data Confidentiality and Subject Privacy

- **Describe how confidentiality will be maintained by providing details about the storage facility, duration of storage, data destruction method, and persons with access to the data.** The investigators will take all possible steps to maintain confidentiality during the study. In order to hold information confidential and secure, only the researchers and research assistant will have access to and/or handle the individual data. Data will be stored on a password protected hard drive in the locked motor speech laboratory. In order to inform future research procedures on development

of motor speech skills, data will be maintained for 10 years prior to being deleted from the hard drive storage. Only the principal investigator and approved research assistants will have access to the data.

- **How will subject privacy be maintained during recruitment, data collection and data analysis?** No information will be released regarding the participant's identity during this study. Participants will be recruited during clinical visits and contact will be made in a confidential clinical setting. Only those with legal access to diagnostic information will participate in recruitment and referral. Data collection and analysis will occur in the ECU motor speech laboratory specifically designated for this purpose. This laboratory is located in a private suite of research laboratories. Video recordings of participants will only be used for analysis of the data and reported in summary format only. If video images are reported in future presentations or publication of the data, permission to do so will be obtained from the participants and legal representatives.
  The confidentiality of the participants will be ensured using the following methods :
    - Birth dates of participants are requested to calculate the age. Neither the name of the respondent nor the actual dates of birth will be retained following completion of data collection.

- **If the participants' data or samples will be used for future research, describe how their privacy will be protected?** Although there are no current plans, if the data is used for future research, participants will be re-consented. In the existing consent form, there is an item where the participant may give permission for future contact.
    - Upon receipt, the data from each respondent will be recorded with coded identifiers only and will be maintained on the hard disk of the principal investigator's computer, located in the ECU motor speech disorders laboratory. Access to this computer is limited to the principal investigator through password security.
    - Participants will be identified by code rather than name on all the research materials, they will not be individually identified in any reports,
    - Data and any lists linking names to identification numbers will be locked in a file cabinet in the primary investigator's office.
    - Only the researchers will have access to the data obtained from each subject.

- **Describe any additional safeguards in place to manage illegal, significantly intimate or potentially embarrassing information gathered in this research study.** Data collection for this study involves participants producing a series of proscribed spoken stimuli. Because of this, it is highly unlikely that information gathered during this study will result in illegal, intimate, or embarrassing information; however, investigators will follow regulatory steps in reporting any illegal activity and will maintain confidentiality regarding any intimate or potentially embarrassing information.

- **Include steps to handle information that requires mandatory reporting to officials, for example physical abuse, emotional abuse or health problems.** Investigators will follow regulatory steps in reporting any abuse or health problems to the proper authorities for investigation.

- **If the research study involves HIV testing, describe the plans for pre/post-test counseling and other related considerations.** N/A

8. Obtaining Consent or Parental Permission

- **Describe the consent process, including members of the research team that will be obtaining informed consent from study participants.** The principal investigator will be responsible for gaining consent from the participant's legal representative, as well as assent from the participant. Participants will be given as much time as needed to fully comprehend and ask any questions regarding the consent forms. The researchers will explain each item on the consent form separately, in order to explain or clarify any vague or misunderstood information. The researchers will also ask questions regarding the information contained in the consent to ensure that the participant fully understands the research study.
- **Describe the setting in which the consent will be obtained.** Individuals interested in participating will be asked to schedule a face-to-face meeting with the investigators in a quiet, private room in the ECU Speech-Language-Hearing clinic, at their home, or preferred location conducive to quiet, confidential discussion. At that session, the research project and the consent, as well as the child's assent will be detailed.
- **Describe the process to minimize undue influence and coercion during the consent process.** Participant and guardian will be informed regarding their right to discontinue participation or withdraw consent at any time prior to or during data collection without any repercussions from the researchers or the ECU Speech-Language-Hearing clinic. The participants will be given the opportunity to ask questions regarding the consent and/or the assent form and if wanted, the participants may take the consent/assent form home with them to discuss with personal confidants and return it at a later date.
- **Outline procedures for obtaining informed consent from participants with limited or low literacy.** Our procedures for consent allow for limited literacy because the researchers will read and discuss each item on the consent form with the potential participant. Because we expect participants to have limited literacy due to their developmental age, we have written the consent forms, and particularly the assent forms, in simple, easy to understand language.
- **Describe the process for determining cognitive impairment or other conditions that may make a participant more vulnerable.** The researcher will ask the legal representative for any medical/educational history of cognitive impairment prior to enrollment in the study and will informally assess the participant's general cognitive abilities by comprehension and ability to perform tasks presented.
- **Describe the process for identifying the legally authorized representative and the process to debrief and subsequently obtain consent from the study participant, when feasible.** N/A

9. Minor Assent Related Issues

- **Describe the assent processes given the range of ages intended for this research study.** The minor participants will be given as much time as needed to fully comprehend and ask any questions regarding the separate assent form. The research team will explain each item on the assent form separately in order to explain or clarify any vague or

misunderstood information. The research team will also ask the participant questions in order to ensure the participant understands the research study.

- **If a separate assent is not being used, how will assent be documented?** N/A
- **How will custody changes during participation in the study be determined?** The legal guardian will re-consent for the minor's participation.
- **Describe the processes as required for enrolling wards of the state if they are a target population for this study. Note : If a child becomes a ward of the state, the IRB must be notified immediately to seek advice on further protections that may be required.** N/A

1. Background

a. Describe the current state of knowledge surrounding the research questions to be addressed in this study.

Historically controversial, the definition of CAS has leveled recently as a neurologically based motor planning speech sound production disorder. Exactly how and what differs in the speech production of children with CAS from typically developing children has not. Early information on the consistency of lip and jaw movement during production of repeated utterances for children, TD and CAS, is not available. Currently, CAS is regarded as "a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (American Speech-Language-Hearing Association, 2007)." Although most professionals agree that CAS is a motor planning problem that originates in the brain, little is known regarding the origin and nature of CAS (Shriberg, 1997). Because the literature identifies this particular group of children with varied characteristics, research questions are designed to address different aspects associated with CAS. Four main areas of concern are typically identified with CAS. These center on the 1) speech sound disorder (Shriberg, Campbell, Karlsson, Brown, McSweeny, & Nadler, 2003; Crary, 1993; Hall, Jordan, & Robin, 1993; Caruso & Strand, 1999), 2) movement (for speech and oral non-speech) (McNeil, Robin, & Schmidt, 1997; Strand, 2003), 3) medical features; and finally, 4) a genetic foundation (Fisher, 2005; Fisher, Lai, & Monaco, 2003; Vargha-Khadem, 2003) has been implicated. This preliminary project addresses (1) speech sound disorder and (2) movement for speech production. Acoustic and perceptual characteristics are both implicated in CAS, including issues associated with speaking rate, timing, and prosody. In addition, numerous reports of increased difficulty, particularly with sequencing sounds, with greater length and complexity of utterances are present in the literature. More recent research has focused on articulation accuracy, examining consonant and vowel error patterns, impaired intelligibility, and inconsistency of productions.

b. Describe the uncertainty to be addressed by this research study (research question).

At a 2002 research symposium held primarily for the purpose of focusing research on Childhood Apraxia of Speech (CAS), Campbell (2003) identified a desperate need for new diagnostic measures that are both reliable and conceptually valid for classification of children with CAS. The goal of this clinical research proposal is to determine whether speech characteristics can be identified that differentiate children with CAS from typically developing children without speech sound disorders. These characteristics must

meet appropriate levels of sensitivity and specificity to be appropriate for clinical intervention and research.

c. Describe the rationale for the type of research design chosen for this study. Once identified, the differential features will then be used to complete studies designed to determine possible physiologic/genetic etiologies and develop targeted interventions. One goal of this project is to move toward a common speech protocol for use by investigators that might also readily be translated directly for use in clinical interventions. Shriberg, Davis, Tomblin, McSweeny, Karlsson, & Scheer (2005) outline an emerging strategy for identifying diagnostic and phenotypic markers of genetically transmitted speech delay; which they hypothesize as the most common subtype of child speech sound disorders from unknown cause. Their strategy involves examination of familial aggregation, perceptual measures, and acoustic analyses to determine a phenotypic characterization. For this project, kinematic, acoustic, and aerodynamic analyses will provide insight into the rate, movement extent & trajectory, placements, and abnormalities in speech production in both a typically developing child and one with CAS.

Sources :

American Speech-Language-Hearing Association. (2007). Childhood Apraixia of Speech. *Technical Report* , 1-74.

Betz, S., & Stoel-Gammon, C. (2005). Measuring articulatory error and consistency in children with developmental apraxia of speech. *Clinical Linguistics and Phonetics, 1*, 53-66.

Bishop, DVM. (2002). Motor immaturity and specific speech and language impairment : Evidence for a common genetic basis. *American Journal of Medical Genetics (Neuropsychiatric Genetics), 114*, 56-63.

Clark, HM., Robin, DA., McCullagh, G., & Schmidt, RA. (2001). Motor control in children and adults during a non-speech oral task. *Journal of Speech-Language-Hearing Research, 44*, 1015-1025.

Davis, B. (2003). Patterns of vowel production in childhood apraxia of speech. In (L Shriberg & T. Campbell, Eds.) *Proceedings Of The 2002 Childhood Apraxia Of Speech Research Symposium*, Carlsbad, CA : The Hendrix Foundation, pp. 49-56.

Dromey, C. & Benson, A. (2003). Effects of concurrent motor, linguistic, or cognitive tasks on specific motor performance. *Journal of Speech, Language, and Hearing Research, 46*, 1234-1246.

Goffman, L., & Malin, C. (1999). Metrical effects on speech movements in children and adults. *Journal of Speech, Language, and Hearing Research, 42*, 1003-1015.

Shriberg, L. A. (1997). Childhood Apraxia of Speech : I. Descriptive and Theoretical Perspectives. *Journal of Speech, Language, and Hearing Research* , 273-285.

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Parental/Legal Guardian Informed Consent Form

Title of Research Study

Speech Production Analyses of Children: Characterizing Typical Development versus Childhood Apraxia of Speech

Invitation

Your child, _____, is invited to participate in this research study. The following is provided in order to help you decide whether to allow your child to participate. If you have any questions, please do not hesitate to ask.

Why is your child eligible?

Your child is eligible to participate in this study because he/she has been identified as a child with childhood apraxia of speech (CAS) or typically developing speech (TDS).

What is the purpose of this study?

The purpose of this study is to determine how children use their oral and facial movements to produce speech. We will use high-speed cameras to capture 3D traces of oral and facial movements while your child speaks. Some of the speech samples that your child will produce will be used to measure features to help us determine the consistency of speech movements and any features that may distinguish CAS from TDS. At this time, methods to distinguish the two do not exist.

What does this study involve?

In this study, your child will complete speech tasks similar to those typically completed during speech testing. Specific procedures include the following standard tasks: (a) standard speech-language assessment (articulation, language), (b) social-behavioral interactions, (c) cognitive screening. In addition, your child will complete a test to determine how understandable he/she is when saying individual words and also in conversation. For all procedures, your child will be seated in a comfortable chair, where researchers will slide a small microphone over his/her ear to record speech. After the first testing session, the researchers will place very small (1-2mm) reflective markers (i.e., very small reflective Styrofoam balls) to several places on your child's face (nose, chin, lips, forehead). We will apply double-sided tape used in other medical applications to affix the markers so as to minimize any risk of allergic reaction or discomfort. Removal of the markers will involve less discomfort than is typically associated with removal of a small band-aid. Once the markers are placed, your child will say a variety

Parent/Guardian Initials_____

of speech items as they are recorded using the digital audio recorder, video, and 3D motion analysis cameras.

Examples of the speech tasks include (a) single sounds, (b) syllables, (c) words, (d) nonsense words, and (e) sentences. Your child will to be seen for these scheduled research sessions at your convenience. Although frequency and length of session is determined based on the child's performance and the tasks presented, it is expected that the entire study should be completed in 3 sessions of approximately 20 minutes each.

The researchers will count the number of correct speech attempts during each session. In addition, we will audiotape and videotape so that we may complete analysis of your child's speech productions after the sessions are complete. The researchers will complete all data interpretation and analysis.

What are the possible risks and discomforts your child could experience?
There are no known risks or discomforts your child could experience during this study. No risk or discomfort has previously been documented associated with any of these measures. Participation will require your child to interact in speech sessions of approximately 20 minutes in duration, completing the tasks involved in usual daily interactions.

What are the possible benefits to your child?
Your child will receive a comprehensive speech and language evaluation as a component of this research project. At this time, although we anticipate finding effective ways of testing speech movements, it is possible that there may be no direct benefit to your child from participating in this study.

What are the possible benefits to society?
The knowledge gained from this study may be of value to other children diagnosed with CAS and to professionals who work with them because it will provide information about testing, designing treatment programs and determining the best ways to treat speech sound disorders in children with CAS. This information is currently unavailable.

Parent/Guardian Initials_____

What are the alternatives to participating?
If you decide not to allow your child to participate in this study, s/he will continue in regularly scheduled speech therapy using conventional methods if currently enrolled. If not currently enrolled in speech, this will not influence participation. Any speech services obtained outside of this research project will involve standard assessments without data collection for research purposes.

What are your financial obligations?
You are not financially responsible for the cost of procedures implemented during the conduct of this study. In addition, you will not receive any monetary compensation for your child's participation in this study.

What should you do in case of an emergency?
If your child has a research-related injury or problem, or if your child experiences an adverse reaction, please immediately contact one of the investigators listed at the end of this consent form.

What are your child's rights as a research participant?
Your child has rights as a research participant. The investigators will be available to answer any questions concerning this research, now or in the future. If you have any questions concerning this research, you may contact the Dr. Laura J. Ball, Ph.D., CCC, telephone (252) 744-6147. If you have any questions concerning your child's rights as a research subject, you may contact the East Carolina University Institutional Review Board, telephone (252) 744-2914. If you would like to report objections to this research study, you may call the ECU Director of Research Compliance at phone number 252-328-9473.

How will your child's confidentiality be protected?
The purpose of the information to be gathered for this research study is to better understand speech motion consistency of children. The individuals who will use or disclose your child's identifiable health information for research purposes include Dr. Laura J. Ball, Jennifer Lemkes, and Susanna Skye Lewis (researchers). These individuals will receive your child's identifiable health information for research purposes only. The type of information accessed for this research study includes previous relevant medical, speech, language, and cognitive evaluation results. The information will be used and disclosed in such a way as to protect your child's identity as much as possible; however, confidentiality cannot be absolutely guaranteed. Someone receiving information collected under this Authorization

Parent/Guardian Initials_____

could potentially re-disclose it, and therefore it would no longer be protected under the HIPAA privacy rules (federal rules that govern the use and disclosure of your health information). There is not an expiration date for this Authorization.

Your child may not participate in this study if you do not sign this Authorization form. You may revoke (withdraw) this Authorization by submitting a request in writing to Dr. Laura J. Ball, at 3310U Allied Health Building, East Carolina University, Greenville, NC 27858. However, the research team will be able to use any and all of the information collected prior to your request to withdraw your authorization.

To authorize the use and disclosure of your child's health information for this study in the way that has been described in this form, please sign below and date when you signed this form. A signed copy of this Authorization will be given to you for your records.

What will happen if you decide not to allow your child to participate?

You can decide not to allow your child to participate in this study, or you can withdraw your child from this study at any time. Your decision will not affect your care or your relationship with the investigator(s) or East Carolina University. Your decision will not result in any loss of benefits to which you are otherwise entitled. If any new information develops during the course of this study that may affect your willingness to continue to allow your child to participate, you will be informed at once.

What conflicts of interest exist?

This research study in not funded. Neither the research site, nor Dr. Laura J. Ball, Ph.D., CCC will receive any financial benefit based on the results of this study.

Documentation of Informed Consent

YOU ARE VOLUNTARILY MAKING A DECISION WHETHER OR NOT TO ALLOW YOUR CHILD TO PARTICIPATE IN THIS RESEARCH. YOUR SIGNATURE MEANS THAT YOU HAVE READ AND UNDERSTOOD THE INFORMATION PRESENTED AND DECIDED TO ALLOW YOUR CHILD TO PARTICIPATE. YOUR SIGNATURE ALSO MEANS THAT THE INFORMATION ON THIS CONSENT FORM HAS BEEN FULLY EXPLAINED TO YOU AND YOUR QUESTIONS ABOUT AREAS YOU DID NOT UNDERSTAND HAVE BEEN ANSWERED TO YOUR SATISFACTION. IF YOU THINK OF ANY ADDITIONAL QUESTIONS DURING THE STUDY, YOU SHOULD CONTACT THE INVESTIGATOR(S). YOU WILL BE GIVEN A COPY OF THIS CONSENT FORM.

Parent/Guardian Initials_____

Signature of Parent/Legal Guardian Date Time

Printed Name

MY SIGNATURE AS WITNESS CERTIFIES THAT THE PARTICIPATING CHILD'S PARENT SIGNED THIS CONSENT FORM IN MY PRESENCE AS THEIR VOLUNTARY ACT AND DEED.

Signature of Witness Date Time

I CERTIFY THAT ALL THE ELEMENTS OF INFORMED CONSENT DESCRIBED ON THIS CONSENT FORM HAVE BEEN EXPLAINED FULLY TO THE PARTICIPANT. IN MY JUDGEMENT, THE PARTICIPANT IS VOLUNTARILY AND KNOWINGLY GIVING INFORMED CONSENT AND POSSESSES THE LEGAL CAPACITY TO GIVE INFORMED CONSENT TO PARTICIPATE IN THIS RESEARCH STUDY.

Signature of Investigator Date Time

AUTHORIZED STUDY PERSONNEL

PRINCIPAL INVESTIGATOR:	Laura J. Ball, Ph.D.	Phone: (252) 744-6147
SECONDARY INVESTIGATOR:	Jennifer Lemkes, B.S.	Phone: (828) 551-0826
SECONDARY INVESTIGATOR:	S. Skye Lewis, M.S.	Phone: (252) 744-6124
SECONDARY INVESTIGATOR:	Jonathon Goodwin, B.S.	Phone: (252) 744-6124

Parent/Guardian Initials_____

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Child Assent Form

Title of Research Study

SPEECH PRODUCTION ANALYSES OF CHILDREN: CHARACTERIZING TYPICAL DEVELOPMENT VERSUS CHILDHOOD APRAXIA OF SPEECH

1. We would like to ask you to be in this study. It is about how you move while you are talking.
2. We want you to ask your parents about this before you decide to be in this study or not. We will also ask your parents whether they want you to be in this study.
3. If you have any questions at any time, please ask.
4. In this study, we will try to find out if we can track how you move your mouth and jaw while you say some words and sentences.
5. In this study, you will tell us what you see on a movie screen or repeat some things that we say. We will stick some small silver colored balls on your jaw and face and use some cameras to see your speech movements. You will be able to watch on the computer screen if you want. We will also put a microphone over your ear so we can record your speech at the same time.
6. There is nothing that will hurt. It will be a lot like if you were in speech therapy.
7. This study may help us understand how children talk. What we learn from you being in this study may help children who have speech problems.
8. You do not have to be in this study. If you decide to be in this study, you can stop at any time. Your decision will not change your relationship with anyone at ECU.

UMCIRB
APPROVED
FROM 6.5.09
TO 3.17.10

Participant's Initials_____

Documentation of Assent
YOU ARE DECIDING WHETHER OR NOT TO BE IN THIS STUDY. SIGNING THIS FORM MEANS THAT YOU HAVE AGREED TO BE IN THIS STUDY AFTER HAVING READ AND UNDERSTOOD THIS FORM. YOU AND YOUR PARENTS WILL BE GIVEN A COPY OF THIS FORM TO KEEP.

Signature of Participant	Date	Time

Printed Name

Signature of Investigator	Date	Time

AUTHORIZED STUDY PERSONNEL

PRINCIPAL INVESTIGATOR	Laura J. Ball, Ph.D.	Phone: (252) 744-6147
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FROM 6·5·09
TO 3·17·10

Participant's Initials_____