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COMMUNICATIVE PARTICIPATION IMPROVES FOLLOWING MOTOR SPEECH
PROGRAM TREATMENT IN APRAXIA OF SPEECH

BY

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B.S Communication Sciences and Disorders, University of New Hampshire, 2016

THESIS

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the Requirements for the Degree of

Master of Science
in
Communication Sciences and Disorders

May, 2018

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Dedication

This body of work is dedicated to my parents, Mark and Liza, for providing me with everything I have ever needed to grow and thrive as a person, student, clinician, and researcher. This project is a culmination of what I was fortunate enough to learn because of you.

Acknowledgements

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Emily A. Schultz

University of New Hampshire, May 2018

ABSTRACT

Childhood apraxia of speech (CAS) is a motor speech disorder characterized by increase in segment and intersegment durations (segmentation), equal stress over words and/or sentences, dysprosody, and speech sound distortions. With decreased intelligibility, limited or lack of communicative participation arises from an inability to be understood or lack of confidence in their speech. Establishing communicative participation measurements is integral to generalizing and establishing efficacy of treatment program progress to a child's everyday life. This study observes the communicative participation change of a group of children (n=6) with idiopathic CAS, receiving a new four-week, 16-hour treatment called Treatment for Establishing Motor Programming Organization (TEMPO). Clinically significant changes were seen in communicative participation post TEMPO treatment using the FOCUS-34© parental questionnaire with an average change of 50 points. Specifically, subscales of intelligibility, social/play, independence, and coping/emotional skills were seen as driving components of this change.

Idiopathic childhood apraxia of speech (CAS) is a motor speech disorder that is characterized by increase in segment and intersegment durations (segmentation), equal stress over words and/or sentences, dysprosody, and speech sound distortions with consistency of error type across repeated production of words (McNeil, Robin, & Schmidt, 1997; Wambaugh, Duffy, McNeil, Robin, & Rogers, 2006). In the presence of average intelligence, these attributes prevent effective communication in all areas of daily living. Influencing social interactions and academic development of school age children, parents of children with CAS also report notable concern with ‘clear speech,’ associated with the Body Functions aspect of the World Health Organization’s (WHO) Functioning and Disability domain of the International Classification of Functioning (ICF) (Lewis et al., 2004; Rusiewicz et al., 2017). While perceptual speech intelligibility is one factor influencing communicative participation and quality of interactions, this study aimed to identify further the components of communicative participation in the presence of a motor speech disorder. The purpose of this study was to provide data on communicative participation of children with CAS who have been treated with a novel treatment protocol, Treatment for Establishing Motor Program Organization (TEMPO).

Childhood Apraxia of Speech

Apraxia of Speech (AOS) is a speech motor programming disorder in which a set of processes that translate complex linguistic (phonological) codes into spatial and temporal patterns of muscle contractions for speech production are impaired (McNeil, Robin, & Schmidt, 1997). This results in distorted speech sound production, impaired prosody, and a very slow rate of speech. In adults, this impairment can be due to damage to the parts of the brain that control how muscles move. This damage may be caused by a stroke, traumatic brain injury, dementia, or progressive diseases. Diagnostic assessments of AOS involve perceptual characteristics that

differentiate AOS from phonological impairments. Childhood Apraxia of Speech (CAS) a developmental motor speech disorder, likely neurological in nature (Plante et al., 2018). The core diagnostic features of CAS, as presented by the American Speech -Language Hearing Association (ASHA) are: “(a) inconsistent errors on consonants and vowels in repeated productions of syllables or words, (b) lengthened and disrupted articulatory transitions between sounds and syllables, and (c) inappropriate prosody, especially in the realization of lexical or phrasal stress (i.e. equal stress on syllables and destress of the stressed syllables)” (ASHA, 2007, p.2). While the symptoms of AOS and CAS are similar, CAS manifests in childhood, while AOS is a result of brain injury or damage to areas of the brain controlling motor movements.

While generally accepted, many experts do not consider inconsistency of errors as key to differential diagnosis in CAS. In a recent chapter, Plante, Miller, and Robin (in press) propose that CAS has the same differential criteria used by McNeil et al. (1997). These criteria are the standard used by evidence-based practice committees to identify CAS in research studies for treatments of the disorder (Ballard et al., 2014). These key differential diagnostic features of CAS are prolonged segment (syllables or speech sounds) and intersegment durations now termed “segmentation”, distortions of speech sounds, and abnormalities in prosody (equal stress on words or syllables). Other secondary clinical features that may be present in speech but are non-differential include: articulatory groping, perseverative errors, increasing errors with increasing word lengths, difficulty with the initiation of speech, awareness of speech and being able to self-correct, and periods of error-free speech throughout their utterances or day (McNeil et al., 1997; Plante et al., 2017; Wambaugh et al., 2006a, 2006b). In addition, the criteria that may rule out CAS would include: fast or normal speech rate, normal prosody, lack of segmentation, and sound reversals (efelant vs elephant). These secondary features aid in providing a more descriptive

diagnosis of CAS that differentiates the disorder from a phonological impairment. This difference significantly impacts the type of treatment that will be most effective in treating these children.

Current Treatments of CAS

Treatment for CAS currently requires more frequent and intensive treatment for a longer duration of time (e.g., ASHA, 2007). In fact, Campbell (1999) estimated that children with CAS require over 80% more treatment than do those children with phonological disorders. Additionally, treatment typically does not guarantee the mastery of all speech sound goals. Findings suggest that observable social characteristics of CAS change with age, although some speech sound goals for these children are met (Lewis et al., 2004). Treatment approaches for CAS currently focus on improving speech sound production to improve intelligibility. Current treatment options fall into motor programming approaches, linguistic approaches, sensory cueing approaches, and rhythmic approaches. Motor-programming approaches utilize the principles of motor learning in which many repetitions of speech movements are practiced with consistency to make speech sounds (e.g. Maas et al., 2008). Linguistic approaches focus on CAS as a language learning disorder, in which children are explicitly taught how to make speech sounds (e.g. Velleman, 2003) and rhythmic approaches use intonation patterns such as stress and melody, to improve overall speech function (e.g. MIT; Albert, Sparks, & Helm, 1973). Each of these approaches targets an individual core symptom of the disorder in isolation (ASHA, 2017).

In a systematic review, Murray, McCabe, and Ballard (2014) indicated the need for a high level of scrutiny relative to identification of key diagnostic symptoms in CAS to evaluate a new treatment to assure that the population targeted by the treatment is an accurate representation of the disorder (ASHA, 2007). Central to their review was the need to demonstrate

maintenance and generalization of treatment outcomes. While there is evidence to support speech sound accuracy improving in case studies of treatments of CAS, there are often remedial speech sound errors, segmentation, and prosodic errors. These lasting errors have lasting impacts on social and academic development and how these children interact in everyday life (Rusiewicz et al., 2017). Treating solely speech production in motor speech disorders such as CAS, is not sufficient for children to make progress in their overall functional communication. With improved intelligibility from treatment, it is anticipated that children will more likely communicate with others, be better understood by unfamiliar people, and independently communicate their thoughts and feelings. However, speech sound accuracy is only preliminary to communicating with peers. Due to the motor programming component of CAS, it is imperative to address all three core symptoms of CAS to improve intelligibility as they jointly occur in a child's speech. Remedial perceptual errors (sound stress and segmentation) can impact a social interaction if not treated beyond speech sounds, leading further to social isolation and decreased participation.

Communicative Effectiveness and Participation

The International Classification of Functioning (ICF) is the World Health Organization's (WHO) health framework striving to provide common descriptions and frameworks for professionals to document or measure the health or presence of disability in children, while creating a profile of their abilities (WHO, 2007). The ICF framework has two parts that would involve the language for a speech-language pathologist: Part 1: Functioning and Disability with a component for Body Functions and Body Structure: physiological functions of the body system including their functions (e.g. articulation or speech sound goals) and another for Activities and Participation: 'the execution of a task or action by an individual and the 'involvement in life

situations' respectively (WHO, 2007, pp 129-130). The ICF defines communicative participation as 'communication in life situations where knowledge, information, ideas or feelings are exchanged' (Eadie et al., 2006). Communicative participation is a complex and multi-faceted construct in which more than speech intelligibility is involved, it involves a person's abilities to perform a particular task in a controlled environment such as the therapy room (their Capacity), and their ability to transfer the information and skills to their everyday life (their Performance). In children with CAS, core symptoms include decreased intelligibility, which falls in the Body Functions Domain. The ICF is a dynamic interaction between Body Functions, Activities and Participation, and their personal history where each domain influences one another. With a limitation in the Body Functions domain for CAS, it is anticipated that their capacity to perform activities in a treatment room will be influenced by their decreased intelligibility, which will then result in a decrease in their Participation, or performance of those learned skills in everyday life.

There are limited communicative participation measures currently in the field of speech-language pathology, particularly in accordance with an effective treatment method for CAS (Thomas-Stonell et al., 2009). Teverovsky, Bickel, and Feldman (2007) investigated parental report to describe the characteristics of children with CAS in terms of functional abilities in reference to the ICF Children and Youth (CY) framework. This framework is oriented to a child's functioning while maintaining similar guidelines to the ICF. This study reported that parents observed functional impairments in articulation, fluency and rhythm of speech, temperament, and mental functions of language, all falling within the Body Functions aspect of the ICF. Items additionally fell in the Activities and Participation aspect, which included conversation, discussion, maintaining attention, learning to write, and writing skills (Teverovsky et al., 2007). As a result of these parental reports, four factors were identified in an effort to

functionally characterize children with CAS. The first was Cognitive and Learning problems including Body Functions and Activity aspects of the ICF. This finding paralleled the longitudinal findings of Lewis, Freebairn, Hansen, Iyengar, & Taylor (2004) of pre-school children with CAS, whom eventually developed learning problems related to reading and writing when they reached school-age. Other factors identified by Teverovsky et al., were Social Communication Difficulties, Behavioral Dysregulation, and Oral Motor Problems, suggesting that there are particular areas in CAS that should be addressed and monitored in addition to improved speech sound accuracy.

The most recent parental reports of experiences with their child's diagnosis of CAS have been collected using the Focus on Communication Under Six FOCUS©, a Likert-scale communicative participation questionnaire (Thomas-Stonell et al., 2010). In 2017, Rusiewicz et al., administered the FOCUS© measure to parents to gather a "snapshot" of functional characteristics of CAS in school-aged children. Parents consistently reported they were concerned about their children's ability to produce clear speech, to communicate effectively, and to be understood by others due to their limitations in speech. These results indicate limitations in the Body Functions aspect of the Functioning and Disability domain of the ICF. More specifically, the lowest scores on the FOCUS© included concerns about clear speech, the ability for their children to be understood by unfamiliar adults, communicating independently and effectively, and to be understood for the first time while speaking to peers. In terms of how CAS impacted their children's everyday activities and social interactions, parents reported concerns about their child's intelligibility impacting peer relationships with reliance on their parents as a 'voice', which indicates Body Functions aspects interacting with the Activities and Participation aspect of the ICF.

In both parental observations in the previously mentioned studies, “snapshots” of functioning in CAS in the absence of a specific treatment method were reported. In the presence of remedial speech sound distortions and the concurrent core features of CAS, it is integral to the field of speech-pathology to observe the social and communicative impacts of these errors. Researchers recommend that speech-language pathologists integrate more goals related to participation into their work, which includes the Activities and Participation chapters of the ICF to treat the disorder holistically.

Development of a New Treatment

As mentioned previously, treatments for CAS are limited in their targeting of the core symptoms of the disorder: speech sound accuracy, word stress, and segmentation. A new treatment method, developed by Robin, is the Treatment for Establishing Motor Programming Organization (TEMPO). TEMPO is structured within a motor learning framework (e.g., Schmidt & Lee, 2005; Maas et al., 2008). The primary goal is to focus simultaneously on speech sound accuracy, prosody and lexical stress (Ballard et al., 2010). The intervention targets each of the three distinguishing features of CAS: distortions, segmentation, and equal syllable stress, as participants repeated the production of multisyllabic nonwords (e.g. butiga) and real words, while using natural speech rate (Miller, 2018). The goal of treating all three core features of CAS is to improve perceptive measures of intelligibility.

This treatment is based on a model of motor programming that has provided evidence for the underlying deficit in AOS/CAS. Specifically, apraxia results from impairments in a working memory buffer that stores individual motor plan just prior to execution (Maas et al., 2008). Part of the working memory buffer is a process that concatenates single motor programs (e.g., syllable into larger motor units, such as words). Another process in the working memory buffer

for speech assigns lexical stress to the overall unit. In essence, children with CAS have difficulty producing smooth (non-segmented) speech with accurate lexical stress patterns and sound production. TEMPO targets this impaired process by having subjects practice three syllable non-words at a normal rate, with accurate prosody and sound production. TEMPO is structured around principles of motor learning, which will be discussed further in the methods, to promote long-term retention and generalization to untreated stimuli and environments (e.g. using at home or school, not just the clinic).

Purpose of This Study:

Contributing to the measures of communicative participation in children with CAS, this study aims to observe changes in communicative participation as a result of TEMPO treatment. Three research questions are asked to clarify our understanding of communicative participation in this sample. First, to examine a comparison between our pre-treatment participation data to published data on participation in children with CAS. We hypothesize that our pre-treatment data of will be consistent with the most recent findings of Rusiewicz et al., (2017) and Teverovsky et al., (2008) in that Body Functions and Activities and Participation domains are limited for children with CAS. Second, we aim to determine if communicative participation changes following a new treatment in CAS. We hypothesize that with improvements in intelligibility (Body Functions), communicative participation will also improve post-treatment. If communicative participation does change post-TEMPO treatment, we aim to determine if specific subcomponents of communicative participation changes post-treatment (e.g. independence, coping skills, being understood by an unfamiliar audience). We hypothesize that if there are improvements in overall communicative participation post-treatment, there will be subcomponents of communication functions that improve more than others.

Methods

Participants

The TEMPO study participants consisted of twelve children with mild to severe CAS, diagnosed by expert clinicians based on the presence of the following features: segmentation, consistent error type, distortions, and equal syllable stress (Ballard et al., 2010). Families were recruited through advertisement flyers, contact with local speech-language pathologists, and website advertising. Ages and genders of each participants in addition to language scores are reported in Table 1. Children were native speakers of English and had no concurrent developmental, neurological, or genetic speech disorders. They all had normal hearing, no muscle weakness, or orofacial abnormalities and received speech therapy up to their participation in the study (Miller, 2018). For this current study, the parents and guardians of the children in the TEMPO study were administered a 34-question measure, the Focus on Communication Under – Six (FOCUS-34©). Each of the nine families received a packet in the mail one to three months post-treatment, including two optional FOCUS-34© measures, for both pre- and post-treatment responses. They were instructed to answer the questionnaires retrospectively as they related to their child’s performance before participation in TEMPO and after treatment. Six (n=6) out of the twelve participating families returned their two surveys via anonymous pre-paid postage to the University of New Hampshire. The University of New Hampshire Institutional Review Board approved of these procedures.

		Treatment Group					Control Group					
		04	06	07	08	Group Mean	01	03	12	15	16	Group Mean
	Age	6;11	5;10	7;8	8;4	7;2	6;7	6;11	7;0	7;5	7;1	7;0
	Gender	M	M	M	F		F	F	M	M	M	
CELF-5 Receptive Language	Sentence Comprehension	4	14	2	7	7	10	14	8	8	12	10
	Word Classes	6	9	7	12	9	9	6	8	8	12	9
	Following Directions	6	7	5	12	8	11	7	5	9	9	8
	Index Score	73	100	69	102	86	100	94	80	89	104	93
CELF-5 Expressive Language	Word Structure	5	9	4	6	6	7	10	10	8	7	8
	Formulated Sentences	3	9	6	10	7	13	8	5	7	9	8
	Recalling Sentences	6	6	4	9	6	13	4	9	8	8	8
	Index Score	69	89	70	90	80	106	85	89	87	89	91
Core Language Score		70	96	66	87	80	102	93	87	86	93	92
Language Content Index		72	96	78	110	89	100	84	80	98	100	92

Table 1: TEMPO participant and group characteristics of language scores, age, and gender for both control and treatment groups. Reprinted from "Improvements in speech of children with apraxia: the efficacy of a treatment for establishing motor program organization (TEMPO)" by Hilary Miller (2018).

Intervention: Treatment for Establishing Motor Program Organization (TEMPO)

The intervention was developed to treat CAS targets three core components of the disorder, segmentation, speech sound accuracy, and lexical stress. Treatment took place four days a week for four weeks, each session an average of 45-60 minutes in order to complete at least 100 practice trials per session. TEMPO is structured using all principles of motor learning (PML) which involves a prepractice and practice phase in each treatment session. The main features of PML include factors associated with practice and those associated with feedback. Practice variables include a high number of repetitions (100), random practice and high complexity sounds. Feedback is in the form of knowledge of results and is presented only 60% of the time during practice and a feedback delay schedule is used.

Specific to TEMPO, prepractice involves randomly selecting 10 SW (strong weak) and 10 WS (weak strong) non-word (e.g. tibuga) stimuli from the overall training set. These stimuli

are administered one at a time, with cues and support specific to Knowledge of Performance (KP) and Knowledge of Results (KR) with 100% frequency. When the participant experiences at least 5 independent, correct responses from prepractice, moving on to Practice is warranted. Practice moves through 100 randomly ordered real and pseudo-word stimuli. Each stimulus is modeled, followed by a delayed repetition (3 seconds) by the participant. Only KR feedback is provided during this phase, as clinicians tell the participant if they are correct or incorrect in the areas of smoothness/fluency, rhythm, and sounds. If the participant fails to get any response correct on the first 20 trials, repetition of the Prepractice protocol is required, before completing all Practice stimuli. Data on the children used in this study show TEMPO to be highly effective in improving segmentation, lexical stress, and sound distortions in both treated and untreated stimuli. Reduced segmentation and fewer distortions were observed in both perceptual and acoustic measures, with a strong effect size (Miller et al., 2018). With these improvements in core features of CAS, perceived intelligibility improves as reported by parents of the participants. In addition to being able to quantify these TEMPO results, the impact outside of the treatment room is essential to contributing to treatment efficacy. To generate comprehensive results of TEMPO across day-to-day living, a communicative participation measure is utilized for generalization of these skills to communicative functions.

Choosing a communicative participation measure: FOCUS-34©

To measure communicative participation for the children in TEMPO, the Focus On Communication Under Six – 34© (FOCUS-34©) is administered to parents of the participants one to three months post-treatment (Thomas-Stonell, et al, 2010). Parents are advised to complete each measure as it represented their child’s performance before and after participation in TEMPO. The FOCUS-34© was developed for measuring communicative participation, rooted

in WHO's ICF, specifically in observing a shift from Body Functions into Activities and Participation as a result of speech and language therapy. The FOCUS-34© captures child capacity and performance as it relates to communication (Thomas-Stonell, 2013). Part one of the FOCUS-34© addresses the components of Body Function or Activities/Capacity. These have been grouped together to evaluate the optimal ability a child has to execute a desired task or action in an environment such as a clinic room. Subcomponents of this section include Expressive Language (e.g. "my child talks a lot," "My child can string words together,") Pragmatics (e.g. "my child uses words to ask for things," "my child waits for his/her turn to talk,") and Receptive Language/Attention (e.g. "my child uses communication to solve problems," "my child can concentrate on the task at hand,")

Part two of the measure addresses Participation/Performance to evaluate what the child does in their day-to-day environment outside the therapy room. Subcomponents of this section include Intelligibility (e.g. "my child's speech is clear," and "my child is understood for the first time when talking to adults,") Expressive Language (e.g. "my child can tell adults who do not know my child well about past events," and "my child can talk to other children about what s/he is doing,") Social/Play Skills (e.g. "my child can communicate effectively with adults who know my child well," and "my child will try to carry on a conversation with adults who do not know my child well,") Independence (e.g. "my child can communicate independently," and "my child can communicate independently with adults who do not know my child well,") and Coping/Emotional Skills (e.g. "my child is comfortable when communicating," and "my child is willing to talk to others.")

The FOCUS-34© questionnaire is intended to be completed by parents and speech-language pathologists intermittently throughout speech-language intervention to measure

changes in communicative participation. Each questionnaire includes 34 Likert-scale questions ranging from 1 to 7 (1 = not at all like my child, 7 = exactly like my child.) Each item score is totaled to generate an overall score and additionally, a change score when comparing two FOCUS-34© measures. A total change score ≥ 11 is considered a clinically significant change (Thomas-Stonell et al., 2012). Item

The FOCUS© has a strong content validity, established by comparing FOCUS© scores with the Ages and Stages Questionnaire – Social and Emotional (ASQ-SE) scores, an established measure of social and communicative competence ($p < .01$). The FOCUS-34©, a shorter version of the FOCUS© demonstrated consistent construct validity and reliability as it reflects original FOCUS© scores almost perfectly ($r = .99$). A positive correlation was observed between the two measures which indicate the FOCUS© is in fact, sensitive to changes in communication and participation skills. Internal consistency for the FOCUS-34© continues to be high for scale scores ($\alpha = .98$) and change scores ($\alpha = .93$) which again, are equal to those of the original measure.

Data Analysis

Data is analyzed by overall FOCUS-34© change scores, subcategory analysis, and individual item analysis. Initially, the total pre- and post-FOCUS-34© items are totaled and compared to report significance. Due to a small sample size, this study uses a one-tailed Mann-Whitney U-test, the non-parametric alternative to the independent sample t-test to determine significance between subcomponents of Capacity and Performance domains and for individual items. The criterion set used an a priori alpha level of .05 to test for significance. Based on the sample size, the critical *U*-value for an alpha of .05 and sample size of 6 is a 7. Scores below seven are considered significant at this criterion. The subcategories making up Capacity and

Performance of these total FOCUS-34© scores were then analyzed with a Mann-Whitney U-test ($\alpha = .05$) to observe driving factors toward the overall significance noted. Scores below seven were considered significant at this criterion. Finally, subcategory items were analyzed with the same U-test criterion to qualitatively discuss the questionnaire items in which significant change occurred for the participants.

RESULTS

Overall Score

Overall results for each participant's change in FOCUS-34© scores are outlined in Figure 1. According to average change ($M = 50$, $SD = 37.56$) between pre- and post-FOCUS-34© Total administration scores, five out of six participants resulted in a clinically significant change (Table 2). These results indicate that over the course of receiving TEMPO treatment, there were observable changes in the communicative participation of a majority of the participants, made by their parents or families. One of the participants resulted in most likely not a meaningful change (score ≤ 6), which will be further elaborated in the discussion. This overall change score does not specify in which ways change was observed. Change in overall score does not depend on a change in each category, significant amounts of growth may be observed in one or two categories alone. This study aimed to determine in specificity, the items that described this change in participation, post-treatment. Analyzed in more depth are the subcategory changes in the questionnaire (Intelligibility, Expressive Language, etc.).

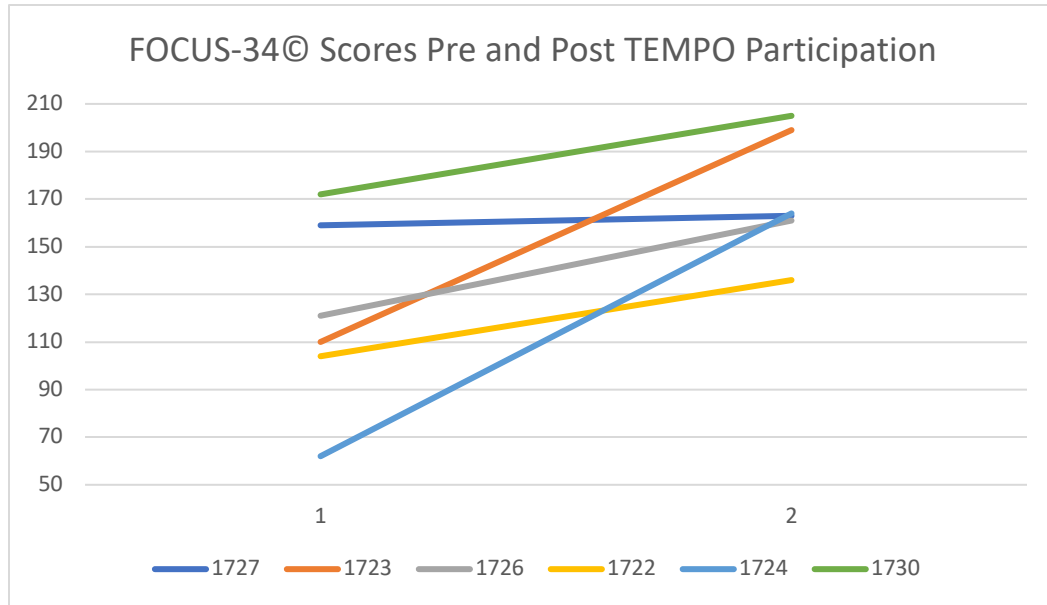


Figure 1: Pre/Post FOCUS-34© scores are outlined above for each participant. X-axis (1) indicated pre-treatment scores and (2) indicate post-treatment scores.

Participant	Pre-Treatment	Post-Treatment	Change Score
1727	159	163	4
1723	110	199	89*
1726	121	161	40*
1722	104	136	32*
1724	62	164	102*
1730	172	205	33*
Average	121.33	171.33	50

Table 2: Pre/Post Total FOCUS-34© scores are outlined above for each participant. The change score indicates the difference between the two administrations. Change scores over 11 indicate a clinically significant change and are indicated with (*).

Subcategory Analysis:

In all eight subcategories changes occurred from pre-and post-treatment. The highest score on the Likert-scale for each subtest is a 7, indicating “exactly like my child” and the lowest score is a 1, indicating “not at all like my child.” Table 3 outlines the subcategories from highest to lowest averages and displays the growth occurring from pre and post administration.

Subcategories of Independence ($M = 2.67$) and Intelligibility ($M=2.8$) were reported as those in which parents indicated “not at all like my child” for a majority of the responses, assuming a

concern with these categories. Following from lowest to higher mean scores include: *Expressive Language Performance* ($M=3.33$), *Expressive Language Capacity* ($M=3.38$), *Receptive Language/Attention* ($M=3.94$), *Pragmatics* ($M=3.95$), *Coping/Emotional* ($M=3.99$), and *Social/Play* ($M=4.07$). Compared to post-treatment scores, categories experiencing the most change were *Independence* ($M=1.91$) and *Intelligibility* ($M=1.9$). The lowest change score was observed in *Receptive Language* ($M=1.06$) and *Social/Play* ($M=0.75$). The other six subcategories had average changes between 1.21-1.44 in their comparisons.

To further distinguish the significance of the changes observed in the subcategories, the Mann-Whitney *U*-test values with the aforementioned criterion set were used to analyze these data (Figure 2). The categories in which significant change in subcategories were observed were: *Intelligibility* ($U=3$), *Social/Play* ($U=7$), *Coping/Emotions* ($U=6$), and *Independence* ($U=4$). These categories align with the Performance/Participation components of the ICF-CY. The *Independent* and *Intelligibility* categories were also those experiencing the most change overall ($M=1.91, 1.9$ respectively) and were also in the categories of most concern for parents at pre-treatment. Subcategories resulting in insignificant changes pre- and post-administration were: *Expressive Language Capacity* ($U=7.5$), *Pragmatics* ($U=7.5$), *Receptive Language* ($U=8$), and *Expressive Language* ($U=8.5$). These subcategories aligned with primarily the Body Functions or Activities/Capacity of the ICF-CY, however, *Expressive Language* ($U=8.5$) is derived from the Performance/Capacity component. These subcategory results exhibit significant change in the Participation domain of the ICF after receiving TEMPO treatment. To further demonstrate what items were driving the changes in these subcategories, the *U*-values for items in each significant category were obtained.

<i>Subcategory</i>	<i>Pre-Treatment</i>	<i>Post-Treatment</i>	<i>Change</i>
Social/Play	4.07	4.82	.75*
Coping/Emotions	3.99	5.23	1.24*
Pragmatics	3.95	5.16	1.21
Receptive Language/Attention	3.94	5	1.06
Expressive Language Capacity	3.38	4.76	1.38
Expressive Language Performance	3.33	4.77	1.44
Intelligibility	2.8	4.7	1.9*
Independence	2.76	4.67	1.91*

Table 3: Average scores (1 = not at all like my child to 7 = exactly like my child) from each subcategory of the FOCUS-34© are illustrated above, from Pre and Post TEMPO treatment. Item values are the average of six participants. Change scores marked with a (*) indicate significant change occurred.

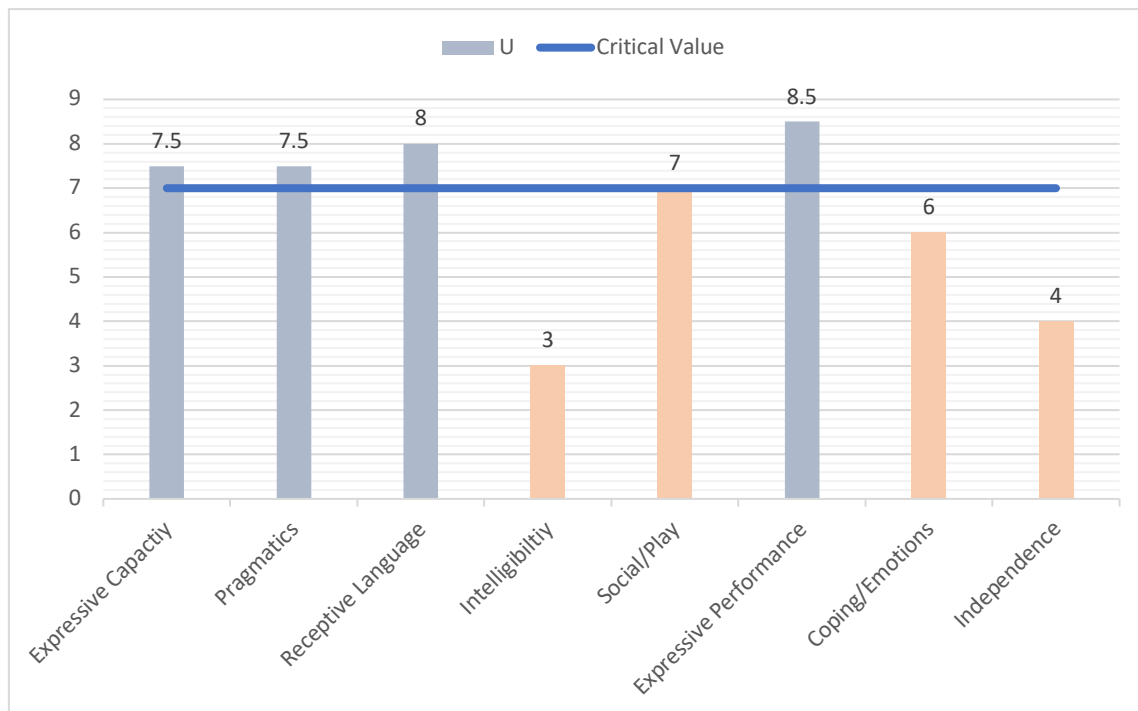


Figure 2: U-Values from Mann-Whitney non-parametric analysis are illustrated above. The blue line indicates the critical value (7) in which items below are considered significant in their change. Categories in orange are those resulting in significant change from pre- and post-treatment.

Item Analysis

In the pre-treatment questionnaire, the lowest scores for individual items parents indicated for their children were within the subcategories of Intelligibility, Expressive Language Capacity and Performance, and Social/Emotional skills. Intelligibility concerns align with the Body Functions domain of the ICF, which includes function of the articulators and motor

components that make up speech (e.g. “my child’s speech is clear”). Expressive Language Capacity is concerned with a child’s ability to use language overall (e.g. “My child uses correct grammar when speaking”) and is in accordance with the Activities Domain of the ICF. Finally, concerns in the areas of Social/Coping skills involve a child’s use of language in social settings, most often with peers (e.g. “my child can communicate with adults who do not know my child well”) which aligns with the Participation domain of the ICF and the performance of “capacity” skills a child has outside of the treatment room.

U-values for individual items in the significant categories of the questionnaire are summarized in Table 4. Out of the four significant subcategories, eleven total items within the questionnaire were deemed significant in their change from pre- and post- treatment measures. The Intelligibility subcategory items included, “My child is understood for the first time when s/he is talking with other children, “my child is understood for the first time when s/he is talking to adults who do not know my child well,” and “My child’s speech is clear.” Social/Play items included: “My child can communicate effectively and carry on conversations with adults who do know my child well,” and “my child can communicate effectively and carry on conversations with adults who do not know my child well”. Two of three Coping/Emotions items were reported significant, “My child is willing to talk to others” and “My child is comfortable communicating,” and all of the Independence items were reportedly significant in their change: “My child can communicate independently,” “my child can communicate independently with other children,” and “my child can communicate independently with other adults who do not know my child well.” The categories in which these items fall, align with the Participation/Performance domain of the ICF. This indicates that pre-treatment concerns of their child’s participation align with the Participation domain, or how their child uses their skills in everyday life. As a result of TEMPO,

which treats the core three symptoms of CAS, the items that improved significantly aligned with Participation and Performance domains. While concerns with Intelligibility are explicitly targeted in treatment, there is a shift of the capacity for skills in “being understood for the first time” towards the use of the acquired skills from treatment.

Mean responses from FOCUS-34© questionnaire	Pre	Post	Change	<i>U</i>	Subcategory
My child can communicate independently with adults who do not know my child well	2.16	4	1.83	3*	Independence
My child uses correct grammar when speaking	2.33	3.33	1		Expressive (C)
My child's speech is clear	2.33	4.33	2	3.5*	Intelligibility
My child is understood for the first time when s/he is talking with other children	2.33	4.5	2.17		Intelligibility
My child is understood for the first time when talking to adults who do not know my child well.	2.33	4.5	2.17	3*	Intelligibility
My child can tell adults who do not know my child about past events	2.67	3.8	1.13		Expressive (P)
My child will try to carry on a conversation with adults who do not know my child well	2.67	4.5	1.83	6*	Social/Play
My child can communicate effectively with adults who do not know my child well	2.67	4.5	1.83	4.5*	Social/Play
My child can communicate effectively with adults who know my child well	3	5.67	2.67	7*	Social/Play

My child speaks in complete sentences	3.16	4.5	1.33		Expressive (C)
My child uses language to communicate new ideas	3.16	5	1.83		Expressive (C)
My child can communicate independently	3.16	4.5	1.33	6.5*	Independence
My child uses new words	3.33	4.67	1.34		Expressive (C)
My child tells stories that make sense	3.33	4.83	1.5		Expressive (P)
My child can communicate independently with other children	3.5	5.5	2	5*	Independence
<hr/>					
My child is confident communicating with adults who do not know my child	3.5	4.5	1	12	Coping/Emotions
My child can string words together	3.67	5.67	2		Expressive (C)
My child conveys his/her ideas with words	3.67	4.83	1.16		Expressive (C)
My child waits for his/her turn to talk	3.67	4	0.33		Pragmatics
My child uses words to ask for things	3.83	5.67	1.84		Pragmatics
My child is comfortable when communicating	3.83	5.3	1.47	5.5*	Coping/Emotions
My child talks a lot	4	5.33	1.33		Expressive (C)
My child can concentrate on the task at hand	4	4.67	0.67		Receptive/Attention
My child can talk to other children about what s/he is doing	4	5.67	1.67		Expressive (P)

My child can be understood by other children	4.16	5.5	1.34	9.5	Intelligibility
My child will ask for things from other children	4.33	5.83	1.5		Pragmatics
My child can communicate effectively with other children	4.33	5.67	1.34	12	Social/Play
My child can respond to questions	4.5	5.5	1		Receptive/ Attention
My child joins in conversations with her/his peers	4.5	6.17	1.67	10.5	Social/Play
My child talks while playing	4.67	6	1.33	7.5	Social/Play
My child is willing to talk to others	4.67	5.83	1.16	5*	Coping/Emotions
My child uses communication to solve problems	4.83	4.83	0		Receptive/ Attention
My child is included in games by other children	4.83	6	1.17	12	Social/Play
My child participates in group activities	4.83	6.17	1.34	10.5	Social/Play

Table 4: Mean responses from each item of the FOCUS-34© pre- and post TEMPO. Scores are organized by Pre-Treatment average responses. Scoring range is 1-7 (1= not at all like my child, 7= exactly like my child. (RL) = receptive language, (C) = capacity, and (P) = performance. The change column is the difference between the two scores. U-Values for items in significant categories are identified with a () if they are considered significant in their change (below critical value 7).*

Children's improvements as reported by parents in free-response question

" My child is not shy and has never lacked confidence, but since receiving this treatment, he has been more likely to engage because he is much clearer and more often understood."

"Adults in his life have all mentioned what a remarkable improvement his speech as made."

"It does not take as long for my child to get their sentences out"

"I not have to interpret my child's message less often when he is speaking to others."

"His teacher can understand him better, he is speaking in a less rushed manner"

"My child is more confident and feels good about himself, he has told me before that he feels dumb. It was heartbreaking to see him struggle and not be able to help."

"I can't imagine what his future would be like without this treatment as he would have totally shut down and not have been able to be his fun loving, amazing spirit that he is."

" I understood my grandson for the first time while talking on the phone with him."

Table 5: Free-responses by parents reporting changes in family, peer, and other social interactions that were not covered on the questionnaire or were elaborated.

Discussion

The children in this study received intervention targeting the three core symptoms of CAS, in which data supports significant changes perceptually, acoustically and via proxy report (Miller, 2018). Across all research questions, we found similarities in pre-treatment data with current literature, improvements in overall changes in participation, and improved subcomponents of communication function. Current literature indicates that communicative participation for children with CAS is impacted in the areas of Body Functions, Activities and Participation domains while concurrently receiving treatments unspecified treatments for CAS

(Rusiewicz et al., 2017; Teverovsky et al., 2008). Our pre-treatment data item analysis continued to support these limitations in the same ICF domains. The Body Functions domain, including intelligibility and speech sound production, are related to the anatomical and physiological components of speech for a child with CAS. TEMPO targeted this domain by training the three core features of segmentation, speech sound distortions, and lexical stress. To investigate the impacts of TEMPO's treatment protocol on communicative participation in CAS, our pre-treatment measures were collected and compared for reference to current literature.

Similarities were found with the top items of concern for both our data and the "snapshot" provided by Rusiewicz et al. (2017). The eight most concerning items for Rusiewicz et al., ($M=2.04$) were the exact items in the top eight concerning items as reported in pre-TEMPO responses ($M=2.43$), with the exception of one item that was not included in the creation of the FOCUS-34© (see Rusiewicz et al., 2017, Table III). The following items were consistent between the two studies, "my child is understood the first time when talking with adults who don't know my child well," "my child can communicate independently with adults who do not know my child well," "my child's speech is clear," "my child can tell adults who do not know my child well about past events," "my child can communicate effectively with adults who do not know my child well," "my child is understood for the first time when she/he is talking with peers," "my child will carry on conversations with adults who do not know my child well," and "my child uses correct grammar when speaking." These items are indicated above the line in Table 4, correlating with the Body Functions and Activities Participations domain of the ICF. Our pre-treatment data correlates with concerns relating to Body Functions, specifically in categories of Intelligibility (e.g. articulation) and Independence (e.g. children not being comfortable or not conveying messages independently.) These findings are also consistent with

Teverovsky et al., (2008) as Body Functions, Activities and Participation were also concerns for parents in this study. In general, the replication of our data to current literature including larger sample sizes, n=201 (2008) and n=40 (2017), strengthens our pre-treatment validity of CAS in the absence of a specific treatment. This validation is useful as the pre-treatment measures were answered retrospectively and the validity of those responses may not be as strong as if they were administered before TEMPO.

With the validity of current literature for our pre-treatment data, results indicated by significant FOCUS-34© change scores demonstrated improvement in communicative participation post-treatment. Our second research question was supported by individual subject data showing that all, but one participant improved in their FOCUS-34© scores post-TEMPO treatment. As a result of participating in TEMPO treatment in which all three core symptoms of CAS are treated, overall communicative participation in the children improved. It was not until further subcomponent and item analyses were completed that improvements in components of communicative functions were observed.

While using current research as a reference for baseline performance of communicative participation in CAS, we question the impact of TEMPO on the Body Functions domain (i.e articulation, intelligibility), leading to a strength in the Activities and Participation Domain. Our hypothesis that areas of specific communicative functions would increase as a result of treatment was supported by significant FOCUS-34© categories. Subcategories with significant changes were *Intelligibility, Social/Play, Coping/Emotions, and Independence* (Figure 2). These four categories align with the Participation/Performance domain of the ICF. With a significant score post-treatment, participants in TEMPO make the most progress in utilizing their skills in their personal and social interactions. This perhaps is due to the increase in skills within Body

Functions which includes targeting the three components of CAS explicitly, shifting to greater skill utilization in Participation or Performance of the skills. Parents report that their child's intelligibility improved, which resulted in higher scores within this subcategory. Examples qualitative reports include, " I understood my grandson for the first time while talking on the phone with him," "adults in his life have all mentioned what a remarkable improvement his speech as made," and "his teacher can understand him better, he is speaking in a less rushed manner". Parental reports indicate close family members and distant adults in a child's life recognizing changes in how well the child is understood post-TEMPO. As intelligibility improves, children become more comfortable utilizing their speech in a natural environment, relying less on someone to translate their messages for them and engaging in communicative opportunities.

Significant items within this category are "my child is understood for the first time when s/he is talking with other children and adults who do not know my child well ($U=3$), and "my child's speech is clear" ($U=3.5$). These items directly reflect the core features of CAS that TEMPO targets. During treatment, participants become aware of their production through consistent self-monitoring of performance on non-word practice. Perhaps through the improvement of perceptual speech skills, the items in the Intelligibility category improved from increased use of these correct productions in the participant's life. By experiencing more communicative success, children understand their messages are being understood at a higher rate than they once were. This logically leads to an improvement in the Independence subcategory of the FOCUS-34©, as the participants understand their autonomy in successful communication.

Significant improvement in the Independence category are related to a child's ability to independently communicate with unfamiliar people and environments without intervention from

parents or caregivers. Parents during pre-treatment responded with lower scores in Intelligibility and Independence, as concerns about their child's ability to produce clear speech and a parent's role as their child's "voice" were expressed. Parents reported their children relied on them to speak for them in social situations, correlating with Personal factors (e.g. feeling frustration or insecurity around speaking) (Rusiewicz et al., 2017). Assuming that an increase in intelligibility generates an awareness of a new expressive opportunities, increased independence and desire to communicate is understandable. This subcategory is integral to beginning to explore social environments and become an independent communicator in school, on the playground, and in society. With the improved confidence in interactions, more communicative opportunities are provided or sought out by these children. Items of the Independence category changing significantly include: "My child can independently communicate", and "my child can independently communicate with other children and adults that know my child well" ($U = 5, 5, 6.5$ respectively). With the decreased reliance on their parents and need for familiar communication partners, these children with CAS experienced increased responsibility for their communication skills, generalizing to their day-to-day lives.

The improvement of the aforementioned subcategories leads to the improvement of Social/Play skills ($U = 7$) where more attempts by the child are made to carry out conversations with unfamiliar communicative partners. Children making more attempts to communicate in their environments and seeking out of interactions is the ultimately evidence of participation. Participation with a wide variety of partners also continues linguistic and cognitive development for these children, as it would occur for their typically developing peers. Items improving within this category include: "My child can communicate effectively and carry on conversations with adults who do know my child well," and "my child can communicate effectively and carry on

conversations with adults who do not know my child well”. Improvements in these items are indications that interactions a child has with a person who does not know them well, is more successful post-treatment. These conversations are ultimately becoming more effective with independence, as parents are no longer interpreters for their children. The improvement in these Social/Play skills promote the use of novel utterances with communication partners that are no longer translated by parents. These skills also continue to promote the use of items in the Independence category, which may continue to strengthen the Participation domain as it pertains to independent interactions. Our pre-treatment data as well as data from Rusiewicz et al. indicate that these particular items were of most concern for parents of children with CAS. A report of significant improvement in these areas is indicative of TEMPO targeting core symptoms that manifest in the most concerning ways for parents.

Further supporting increased confidence in their communication skills, the Coping/Emotions category changed with significance. This category speaks to the child’s performance in feeling comfortable and confident communicating with others. Ultimately, a child’s confidence in communication will allow them to experiment with language, make more attempts at relationships, and extend themselves into learning and academia as their speech is no longer a concern for them. Coping/Emotion skills resulting from an improved intelligibility speak to the impact their speech differences have on their behavioral and emotional reactions to frustrations surrounding speech. Parents report change in performance regarding decreases in frustrations and outbursts for example, "My child is more confident and feels good about himself, he has told me before that he feels dumb. It was heartbreaking to see him struggle and not be able to help." These items speak to the impact outside of the treatment room children are experiencing with CAS. Specific items include, “my child is willing to talk to others” and “My

child is comfortable when communicating” ($U=5$, $U=6$), which were also qualitatively reported by parents reporting, " My child is not shy and has never lacked confidence, but since receiving this treatment, he has been more likely to engage because he is much clearer and more often understood." Reports such as this report a baseline confidence level in their children with CAS. However, confidence does not necessarily relate to initiation or engagement in conversations with peers. As a result of participation in TEMPO, children with confidence are able to eliminate the concerns of their speech and express themselves to communication partners and peers in naturalistic environments.

While the overall communicative participation score for the group ($n=6$) improved significantly, one participant demonstrated not likely a clinically meaningful change (change = 4). While the overall score was not significant, individual categories did improve (Expressive Language Capacity, Receptive Language, Independence, and Coping/Emotional). Within the subcategories in which no change occurred (Intelligibility, Expressive Language Performance) or performance decreased (Social Play, Pragmatics), individual item analysis was informing of alternate changes. Pragmatic items that decreased were those associated with “waiting for their turn to talk” (change = -0.16). As qualitatively reported by parents of this study, these children had “increased confidence in a desire to talk”, and perhaps the hesitancy during conversations was being impacted or eradicated by this new skill. Intelligibility items for this participant did not show signs of change in the overall score (change = 0), while individual items of “being understood for the first time when talking to other children” improved and “can be understood by other children” decreased, equalizing the score. The relationship between these two items suggests an assumption in the perception of other children by observation of parents. The increased intelligibility when speaking “for the first time” to unfamiliar audiences can be

arguably more powerful in creating new relationships and encountering novel environments as children mature. The analysis of this participant individually demonstrates the importance of individual item's to be representative of participation conditions across daily living, to allow for accurate examination of areas for improvement in individual treatment.

Future Directions

These data suggest a necessary direction to investigate efficacy of treatment in motor speech disorders, particularly childhood apraxia of speech. While the population involved in this study was small, thorough evaluation procedures to diagnose CAS were taken to ensure the population fully described the participants. In order to observe a greater treatment impact, investigating a larger population of participants would further validate how communicative participation is impacted by a motor speech disorder such as CAS. Additionally, this study utilized a measure (FOCUS-34©) intended for a population under six. Due to lack of validated measures specifically for communicative participation, this measure was utilized. The results of this study should guide future development of measures that target specific populations of speech disorders, as not all are reflected similarly in participation (Lewis et al., 2004). Results of these measures may aid in creating communicative profiles for children with motor speech disorders, creating a prognosis for participation beyond childhood. Treatment goals utilizing particular treatments such as TEMPO can incorporate goals relating to communicative participation when a foundation of participation skills is established for CAS.

This study also collected data retrospectively for pre-treatment data. Ideally, to gain a more accurate representation of children's current communicative participation before TEMPO treatment, giving parents or guardians the questionnaire before treatment would be more effective. Although our data aligned with that of other parent reports of their children with CAS,

these pre-treatment measures provide baseline measures that are integral to measuring progress. Multiple administrations from the baseline data would also be beneficial to understand the timing of changes in communicative participation and to gain a model describing the components of speech that correlate with participation improvements.

Participation measures are increasingly important for the holistic treatment of children and adults, alike. Eadie et al 2009 indicate “information provided by the person with the health impairment is an essential component of measuring participation because it will provide a unique insight into what the measures currently can’t measure.” Including participant perspective may perhaps be beneficial if older children or adults are recruited for TEMPO treatment. It would be necessary for participation measures to be accommodating of activities and day-to-day events in which communication is crucial. By gaining insight into participation from the participant themselves, a new perspective of the linguistic and cognitive development of these children will emerge. With potential remediation of the core symptoms of CAS, children can contribute to develop similar to their peers, engaging in conversations and academics with new accessibility.

Ideally, more participation measures in the field of speech-language pathology continue to develop. Results from treatment trials such as TEMPO provide a promising future for not only the intelligibility impacts of a motor speech disorder but for the social impacts that may burden children with any speech and language disorder. Measures can be further developed to specifically target disorders by including items sensitive to their experience.

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Appendix A: Institutional Review Board Approval

University of New Hampshire

Research Integrity Services, Service Building
51 College Road, Durham, NH 03824-3585
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IRB #: 6627

Study: TEMPO Treatment Study

Approval Expiration Date: 04-Apr-2018 **Modification Approval Date:** 27-Nov-2017

Modification: Changes per 10/30/17 Memo

The Institutional Review Board for the Protection of Human Subjects in Research (IRB) has reviewed and approved your modification to this study, as indicated above. Further changes in your study must be submitted to the IRB for review and approval prior to implementation.

Approval for this protocol expires on the date indicated above. At the end of the approval period you will be asked to submit a report with regard to the involvement of human subjects in this study. If your study is still active, you may request an extension of IRB approval.

Researchers who conduct studies involving human subjects have responsibilities as outlined in the document, Responsibilities of Directors of Research Studies Involving Human Subjects. This document is available at <http://unh.edu/research/irb-application-resources> or from me.

If you have questions or concerns about your study or this approval, please feel free to contact me at 603-862-2003 or Julie.simpson@unh.edu. Please refer to the IRB # above in all correspondence related to this study. The IRB wishes you success with your research.

For the IRB,



Julie F. Simpson Director

cc: File