EFFECTS OF CHILDHOOD APRAXIA OF SPEECH TREATMENT ON FUNCTIONAL COMMUNICATION OUTCOMES

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ABSTRACT

The goal of this research was to determine if Integral Stimulation (IS), a treatment used for children with childhood apraxia of speech (CAS), resulted in improvements in functional communication outcomes. In previous research, success or progress in CAS treatment focuses on articulatory precision, but this research aimed to look at progress from a more functional standpoint within the context of the World Health Organization's International Classification of Functioning Disability and Health (ICF). The WHO ICF breaks down functioning, and in this case communicative functioning, into components that interact in a nonlinear fashion. These components are "body functions and structures", "activities", and "participation". By looking at measures across these domains, we are able to obtain a more holistic view of the impact of treatment.

The research was completed via a retrospective analysis of data obtained from the families of two children with CAS. As part of a study on the effectiveness of IS treatment, the families completed two indirect measures of communicative function — namely the Focus on Outcomes of Communication Under Six (FOCUS-34) and the Intelligibility in Context Scale (ICS). The goal of these questionnaires is to assess communication in the child's daily interactions with family members, teachers, and peers by using a Likert scale to quantify various aspects of their communication skills. The questionnaires were completed twice before treatment was implemented and one more time at the conclusion of treatment.

The results of the study suggest that broad changes in functional communication outcomes were observed but were not reliably attributable to treatment. However, there is evidence that in a deeper analysis, IS treatment may result in measurable improvements

within specific WHO ICF domains. The pattern of changes was not clearly predictable from changes in speech accuracy, indicating that functional improvements cannot be assumed from accuracy measures and instead must be measured separately. The results indicate that further research is needed in determining these domain-specific functional outcome measures for CAS treatment.

I dedicate this thesis to my mom

Veronica. For her advice, her patience,

her faith, and of course, her red pencils.

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I wish to express my deepest gratitude to my husband Andre, my siblings, and my mother. Thank you all for supporting me every step of the way and instilling in me the confidence and determination to accomplish this work. While my father was not here for this part of my journey, I am forever grateful for his support and faith in me and know that his influence is at the root of this accomplishment. The completion of this research would not have been possible without my family's patience, love, and unrelenting support.

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CHAPTER 1

INTRODUCTION

Childhood Apraxia of Speech (CAS)

Childhood apraxia of speech (CAS) is a pediatric motor speech disorder. According to The American Speech-Language-Hearing Association (ASHA, 2007), CAS is defined as a speech motor planning disorder that results in the inaccurate, inconsistent motor production of speech sounds that cannot be explained by abnormal neuromuscular function. The underlying problem in CAS is a difficulty in planning the movements required for speech production. Three key indicators of a CAS diagnosis are inconsistent consonant and vowel errors, extended or inconsistent transitions between sounds, and difficulties with prosody, in particular lexical and phrasal stress.

Children living with CAS or other speech disorders are presented with additional hardships and obstacles as they navigate social, academic, and eventually professional spaces (Felsenfeld, Broen, & McGue, 1994; Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004; McCormack, McLeod, McAllister, & Harrison, 2010). Felsenfeld et al. provided evidence of the otherwise assumed challenges that these children could face. In their study, they found that the individuals who were diagnosed with a phonological disorder required additional interventions throughout their educational careers, and ultimately received fewer years of education. Similarly, McCormack et al.'s literature review noted that these children not only struggle in academic settings but in their interpersonal skills with families, friends, people of authority, and professional peers.

Sound epidemiological data are not available due to the historical controversy surrounding the existence and diagnosis of CAS. However, using information from

clinics working with preschool aged children, it has been estimated that 1-2 children per one thousand are diagnosed with CAS (Shriberg, Aram, & Kwiatkowski, 1997). Additionally, 3.4%-4.3% of pediatric cases of speech disorders involve CAS in some capacity (Delaney & Kent, 2004; as cited in ASHA, 2007). According to ASHA's 2018 SLP School Survey (ASHA, 2018), 60% of speech-language pathologists (SLPs) regularly work with children with a CAS diagnosis and approximately three children per caseload have a CAS diagnosis. Given the large, life-changing impact of CAS, it is important to provide effective treatment for these children.

Treatment Approaches for CAS

Children with CAS often require extensive treatment and often make little or slow progress (ASHA, 2007; Campbell, 1999; Shriberg et al., 1997). Several treatment approaches exist and have some evidence to demonstrate their efficacy (see Maas et al., 2014; Murray et al., 2014 for reviews). According to ASHA, regardless of strategy, treatment must be frequent, intense and ongoing in order for children with CAS to show improvement.

Murray, McCabe, and Ballard (2014) divided treatment approaches for CAS into two broad categories, namely motor-based approaches such as Dynamic Temporal and Tactile Cueing (DTTC; Strand, Stoeckel, & Baas, 2006) and Rapid Syllable Transition (ReST) treatment (Ballard, Robin, McCabe, & McDonald, 2010; Murray, McCabe, & Ballard, 2015) and language-based approaches such as the Integrated Phonological Awareness treatment (Moriarty & Gillon, 2006).

The Dynamic Temporal and Tactile Cueing (DTTC; Strand & Debertine, 2000; Strand et al., 2006) approach, which is a form of integral stimulation treatment, combines

simultaneous imitation and tactile cues during drill activities to address targeted real words and sound clusters based on individual client needs. The focus of treatment is on movements needed for speech, rather than sound classes. In the beginning of this treatment method, the clinician focuses on sound production by encouraging slow, elongated utterances with the child. The clinician may incorporate gestures and tactile cues to assist in the correct formation of the articulatory positions. As the child progresses through treatment, the clinician will gradually require faster production to work toward a natural production of the utterance. Eventually the simultaneous practice is replaced with delayed repetition and spontaneous production (Strand et al., 2006). In studying four children with CAS treated over the course of four to six weeks, DTTC treatment resulted in significant improvement in the targeted utterances. The success of these children appeared dependent on commitment to repeated practice and movement gestures (Strand et al., 2006).

Rapid Syllable Transition Treatment (ReST; Ballard et al., 2010; Murray et al., 2015) focuses on drilling pseudowords in order to tackle impaired prosody and articulation. In the "prepractice" portion of treatment, the clinician works to ensure proper production of the nonwords and then releases the client to the practice portion. Then the client completes at least one hundred trials presented in random order with delayed feedback. As a group, the fourteen children with CAS who participated in this treatment study showed a treatment effect for generalization and retention compared to the control group (Murray et al., 2015).

Integrated Phonological Awareness (IPA) focuses on phonological awareness improvement in four tasks—distinguishing phonemes in isolation, word-final and word-

initial environments, segmentation and blending, and manipulation (Moriarty & Gillon, 2006). These tasks are integrated into clinician-chosen activities or games that are designed to keep the client interested and engaged. The progress of three children who had received the treatment was analyzed through their percent phonemes correct in the various tasks. Two of the three participants showed significant improvements in targeted speech, reading tasks, and phoneme segmentation and manipulation for both trained and untrained tasks (Moriarty & Gillon, 2006). Thus, this study suggested that significant improvement can be made in a relatively short amount of time for both targeted and non-targeted control words.

Functional Outcome Measures

Although there is some evidence for each of these treatment approaches, a major limitation of the evidence base to date is that the outcome measures focus heavily on the impairment level rather than on more functional measures (Kearney et al., 2015). More functional measures are intelligibility and how well a child can interact with family and peers. Intelligibility refers to the ability of a listener to understand the speaker's spoken message; a child's success in interacting with others is a subjective assessment that combines expressive language, receptive language, and social-emotional factors.

The World Health Organization's International classification of functioning, disability and health (ICF) presents a model to illustrate the many components that play a role in functionality. All components interact in a nonlinear fashion with each one impacting the others. "Body functions and structures" is the term used to encompass the functions along with the physical parts of the body that are involved. "Activities" are defined as the implementation of a functional task that requires these body structures and

functions. "Participation" is the ability to partake in an activity in daily life. "Context Factors" include both personal and environmental factors that may impact the previously stated components (WHO, 2007). For the components of activity and participation, there are two qualifiers – capacity and performance. Capacity refers to "the highest probable level of functioning" (WHO, 2007) in a controlled setting (i.e. a speech clinic). Performance refers to how an individual functions in his/her everyday environment (i.e., the classroom). An impairment at any of these levels may result in disability with a variety of implications.

In terms of CAS, the impairment occurs during motor planning (body function), which impacts both the activity and participation levels of the WHO ICF model. The activity related to CAS is speaking to communicate, and participation is using this ability to speak in order to accomplish daily tasks and successful communication. Previous studies on CAS treatment primarily focus on the impairment level by looking at the accuracy of speech production and articulatory movements (Kearney et al., 2015; Murray & Iuzzini-Seigel, 2017). A common, but usually implicit assumption is that improving the movements and sound production will result in improved intelligibility (a measure at the activity level) and therefore, improved participation in life experiences. However, by studying the impairment level alone, there is less focus on the activity and participation levels which may in fact be a stronger, more meaningful reflection of treatment outcomes in children with a speech sound disorder. If the goal of CAS treatment is to improve communication and participation in daily experiences, then the activity and participation levels should be analyzed as well. By studying the perceived intelligibility and communication outcomes in daily interactions, the analysis of treatment expands beyond

the impairment level and into the activity and participation levels, and in turn presents a more holistic view of the functionality provided by treatment.

Measures of Intelligibility and Communication Outcomes

Intelligibility can be studied both directly and indirectly through a variety of measures. Indirect intelligibility refers to the degree in which a speaker is understood as judged based on a rating scale. The rating is commonly completed by familiar listeners such as parents and teachers and is meant to reflect the overall intelligibility of the child in various settings over broad timespans. Direct intelligibility looks at specific samples of speech in which the intended utterance is known and can be used as a comparison to the child's production.

The Intelligibility in Context Scale (ICS; McLeod, Harrison, & McCormack, 2012) was developed as a measurement of indirect intelligibility for children with speech sound disorders. Parents or caregivers complete the brief questionnaire as it relates to their child's speech over the past month. The five-point Likert scale provides a numerical representation of how frequently the child is understood in a variety of settings with various communication partners. By looking at speech in daily life there is less emphasis placed on the specific sounds and movements produced. Instead, the ICS provides a framework for assessing overall communication efficacy and quantifies its impact on the participation level of the ICF model.

In a study looking at the validity and reliability of ICS, 120 children were assessed using the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, Hua, Crosbie, Holm & Ozanne, 2002; as cited in McLeod et al, 2012). None of the children were diagnosed with a speech sound disorder, but parents and teachers reported

concerns with speech production in 109 of the total participants. The DEAP results were compared with the parent-completed ICS to determine the validity of the assessment. Analysis showed that the mean score of the ICS was successful in differentiating the children who were identified to have speech production concerns from those who were not. It is important to note that parents and teachers reported the concerns regarding the children's speech production, which could impact the manner in which the ICS was completed. Because they already had reported these concerns, their perspective and opinions during the completion of the ICS could be skewed compared to scenarios in which parents complete the ICS in a more blinded manner. The study also concluded that the ICS has criterion validity by comparing the phonology subtests of the DEAP to the results of the ICS.

In addition to the assessment's strong validity and reliability, the simplicity and lack of financial demand make this a desirable tool for assessing intelligibility. The questionnaire consists of only seven questions and requires minimal effort from the person completing it. The method of scoring is straightforward and simple. Additionally, the assessment has been translated into over 50 different languages with both monolingual and bilingual versions and is available online free of charge (McLeod, 2019). Although the ICS is based on subjective judgments and has limited precision, it has been shown to be a valuable, reliable and valid intelligibility assessment.

The Focus on Outcomes of Communication Under Six (FOCUS; Thomas-Stonell, N. L., Oddson, B., Robertson, B., & Rosenbaum, P. L., 2010) is another tool used to indirectly measure speech intelligibility and communicative participation in children. The assessment was developed to analyze and quantify the components of the WHO's ICF

model to determine the impact of speech and language therapy on communicative activity, participation, and personal factors. Parents, caregivers, or speech-language pathologists complete the 50-item questionnaire, using a 7-point Likert rating to assess how the child uses language and how communication impacts interactions in peer-based settings. Sample questions include “my child’s speech is clear”, “my child can communicate independently with other children”, and “my child makes friends easily”. The assessment takes, on average 20 minutes to complete.

In developing the FOCUS, caregivers and clinicians from 210 children who received speech therapy, provided statements regarding changes that had been observed in the child. The children involved had varying diagnoses with 41% of them having a developmental speech sound disorder. Following the initial creation of test items from these observational comments, an additional 165 families participated across a three-phase study to evaluate consistency, redundancy, reliability, and validity of the assessment tool. The FOCUS originally consisted of 103 items but was ultimately edited to 50 items to reduce redundancy and account for observations related to younger children. Results of the 3-phase study revealed construct validity in its sensitivity to changes in children’s communication and high internal consistency (Thomas-Stonell, Oddson, Robertson, & Rosenbaum, 2010).

In addition to its high construct validity and consistency, the FOCUS is a user-friendly, straightforward assessment that can be used in a variety of situations to assess the impact of speech and language therapy. The variety of questions used provide insight into the child’s communication and how they participate in settings beyond a speech and language therapy session.

More recently, a shorter version with 34 items, the FOCUS-34, has been developed for more efficient clinical use (Oddson et al., 2019). This shorter version has been shown to have comparable validity to the 50-item FOCUS and can also be used to detect change over time, including in response to treatment (Oddson et al., 2019). Both the full FOCUS and the FOCUS-34 also offer information to determine profiles according to the WHO ICF model. The manual categorizes question items into the groups Capacity (which includes the Body Functions and Activity levels) and Performance (which corresponds roughly to the Participation level), so that change in different ICF domains can be charted. Although body function and capacity are not grouped in such a manner in the WHO ICF model, in order to follow the previously determined scoring profiles described by the FOCUS manual, this grouping was maintained for the present study.

In order to assess communicative changes within the context of the WHO-ICF model, the questions from the FOCUS-34 were related to the ICF domains they reflected and grouped into the categories of "capacity" and "performance" (Thomas-Stonell, Oddson, Robertson, & Rosenbaum, 2010). As previously discussed, these are two qualifiers for the ICF components of activity and participation. Due to the dynamic interaction of the ICF components, measures of activity and participation inherently encompass body function and structure (i.e. movements and sound production). By analyzing these two qualifiers, we are able to see how treatment impacted the child's ability to execute the task of speech (i.e. changes in movements/sound production, intelligibility, and activity) and how treatment impacted the child's ability to *use* speech within a real-world context (i.e. participation and context factors).

As previously mentioned, intelligibility has not been utilized frequently as a measurement of treatment efficacy in children with CAS. To the best of our knowledge, no published studies have used either the FOCUS or ICS intelligibility as an outcome measure in treatment for CAS, with the exception of Namasivayam et al. (2015) who used the FOCUS as one of their outcome measures. Namasivayam et al. (2015) reported that children with CAS who received more, and more intensive, treatment (20 sessions over 10 weeks) showed significant gains on the FOCUS total score, whereas children with CAS who received less, and less intensive, treatment (10 sessions over 10 weeks) did not. Namasivayam et al. (2015) did not report changes in different ICF domains separately.

The Present Study

Given the information and previous research conducted, the present study will examine the functional communication outcomes of two children with CAS who completed a treatment study using Integral Stimulation (IS). In this treatment, different sets of items were paired with three different conditions—audio only, visual only, and audio-visual, plus an untreated control condition. The findings from speech accuracy outcomes (a Body Function domain) were reported elsewhere (Condoluci, 2020) and will not be discussed further here. In the present study, the FOCUS-34 and the ICS were used to investigate possible effects of the overall treatment on these more functional parent-rated outcome measures related to communicative function and participation.

Figure 1: Treatment Timeline; *Tx 1 = Treatment phase 1; Tx 2 = Treatment phase 2; M1 = maintenance phase 1; M2 = maintenance phase 2*

	Assessment				**Baseline**				**Tx 1**				**M1**		**Tx 2**				**M2**			
Weeks	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22
FOCUS	T1							T2											T3			
ICS	T1							T2											T3			

The primary question of this study is whether or not treatment results in generalized improvements in intelligibility and communication outcomes. We will address this question by examining children's performance in daily life, as rated by parents on two questionnaires (ICS and FOCUS-34) completed at T1, T2, and T3. A secondary question is whether the treatment has a differential impact on different WHO ICF domains, as captured by the FOCUS-34. We will address this question by examining changes on different WHO ICF domains separately.

CHAPTER 2

METHODS

Speakers

The FOCUS-34 and ICS data used to examine any changes in functional communication were obtained from the families of two children with CAS (see Table 1), who completed the questionnaires over the course of sixteen weeks. The speech accuracy data from this treatment study were presented separately elsewhere (Condoluci, 2020). The children were first evaluated by an experienced SLP who specializes in CAS to confirm their diagnosis. All evaluation and treatment sessions were completed at the Speech, Language, and Brain Lab at Temple University with the approval of the Temple University Institutional Review Board. The parents provided written consent for their children's participation in the study and the children both provided assent.

Treatment Context

The focus of the present study is on the intelligibility and communication outcomes, but a brief description of the treatment design is provided to explain some relevant context. The data were obtained from a single-case experimental design study using an alternating treatments design. Over the course of treatment, the children practiced speech targets in three different conditions—audio only, visual only, and audio-visual, referring to the type of model provided by the clinician during treatment. Once per week, the children completed a probe task at the beginning of the session, in which they repeated the target words/phrases as well as untreated words/phrases in random order after the clinician's model. Sets were matched for word length, interest area, and number of bilabial and labiodental consonants.

Table 1

Participant Information

Information	Participant 001	Participant 003
Age	4;4	4;10
Sex	Male	Male
GFTA-3[a]		
Raw Score	70	58
Standard Score	71	67
Percentile Rank	3rd	1st
EVT-2 (Form B)[b]		
Raw Score	47	40
Standard Score	92	80
Percentile Rank	30th	9th
PPVT-4 (Form B)[c]		
Raw Score	54	62
Standard Score	88	88
Percentile Rank	21st	21st
R&K[d]		
Structural Score	24/24 (100%)	23/24 (96%)
Functional Score	71/102 (70%)	88/102 (86%)
RIAS[e]		
T-Score Sum	89	104
Percentile Rank	30th	63rd
DEAP[f]		
Word Inconsistency Score	76%	67%

[a] GFTA-3: Goldman-Fristoe Test of Articulation 3 (Goldman & Fristoe, 2015)
[b] EVT-2: Expressive Vocabulary Test, Second Edition (Williams, 2007)
[c] PPVT-4: Peabody Picture Vocabulary Test, Fourth Edition (Dunn & Dunn, 2007)
[d] R&K: Oral mechanism exam from Robbins & Klee (1987) protocol
[e] RIAS: Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003)
[f] DEAP: Diagnostic Evaluation of Articulation and Phonology (Dodd et al., 2006)

Treatment was conducted twice weekly for one-hour sessions over eight weeks (i.e. 16 hours of treatment) by a certified and licensed speech-language pathologist and involved a version of integral stimulation treatment. All items were probed during

baseline, during treatment, and following treatment (see Figure 1). During treatment phases, probes were administered at the beginning of the session, before treatment. Probe procedures involved the child repeating the items after the clinician. Probes included items from all sets (treated and untreated) in a different random order each probe. The treating clinician administered the probes. No feedback on accuracy was provided during probes.

Data Sources

Twice before initiation of treatment (T1, T2) and once after completion of treatment (T3), the same parent completed two questionnaires to provide information about their child's communicative function (see Figure 1). In particular, parents completed the ICS and the FOCUS-34. The ICS primarily captures comprehensibility in the child's daily life (the degree to which listeners can understand the child's speech in a given context). The FOCUS-34 captures communicative function more generally, including communicative ability (capacity), communicative function and participation (performance).

Data Analysis

In order to determine the effect of treatment on intelligibility and communicative participation in the two children studied, we will analyze the ICS total scores and FOCUS-34 total scores.

To answer the primary question (i.e., did the treatment result in generalized improvements in intelligibility and communicative participation?), data from the ICS and FOCUS-34 will be plotted and analyzed visually for any changes across time points. Given that the treatment occurred between T2 and T3, we expect greater improvement

between these two time points than between T1 and T2, during which interval the children did not receive treatment.

To address the secondary research question, we will plot and analyze the FOCUS-34 sub scores pertaining to the WHO ICF domains, in order to determine whether some domains demonstrate a greater response to treatment than others.

CHAPTER 3

RESULTS

Overview

The data from both the ICS and FOCUS for each child were collected and analyzed visually and quantitatively across the three timepoints. To address Research Question 1, the ICS and FOCUS scores were analyzed to determine if the intervention, occurring between timepoint two (T2) and three (T3), resulted in greater change than from timepoint one (T1) to T2, where intervention had not yet begun. Similarly, to address Research Question 2, specific sub profile scores from the FOCUS-34, corresponding to Capacity and Performance aspects of the WHO ICF model, were plotted across timepoints to explore any potential differential effects of treatment on these aspects of function.

Research Question 1: Generalized Effects of Treatment

ICS scores for Participant 001 and Participant 003 are presented in Figures 2 and 3, respectively. FOCUS-34 scores for each participant are presented in Figures 4 and 5 Complete data tables for each child are presented in APPENDIX.

Figure 2: Participant 001 ICS Scores

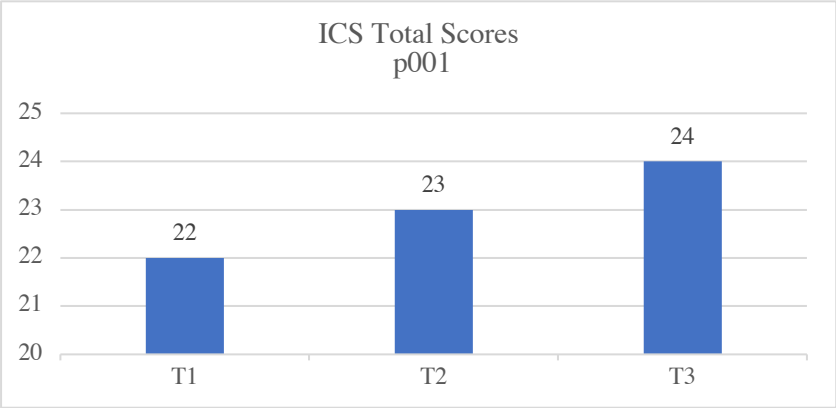


Figure 3: Participant 003 ICS Scores

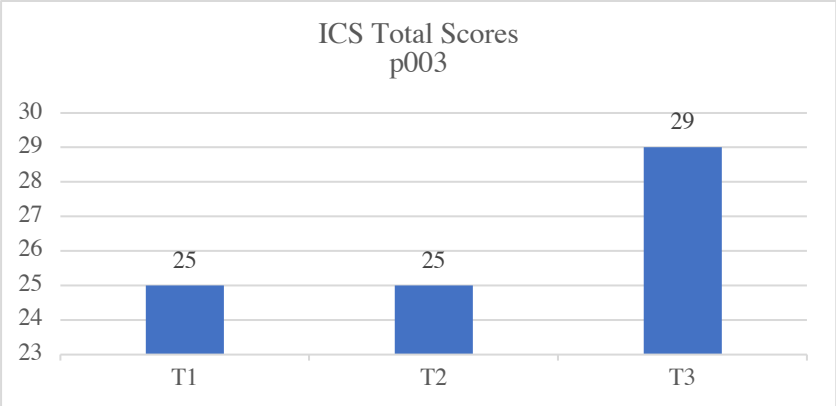


Figure 4: Participant 001 FOCUS-34

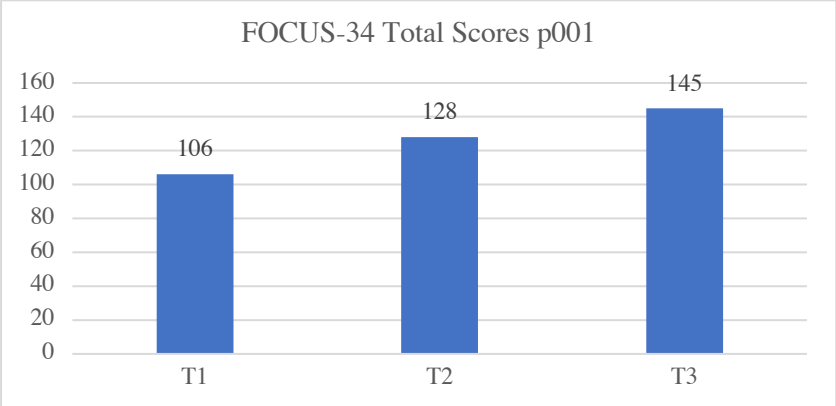
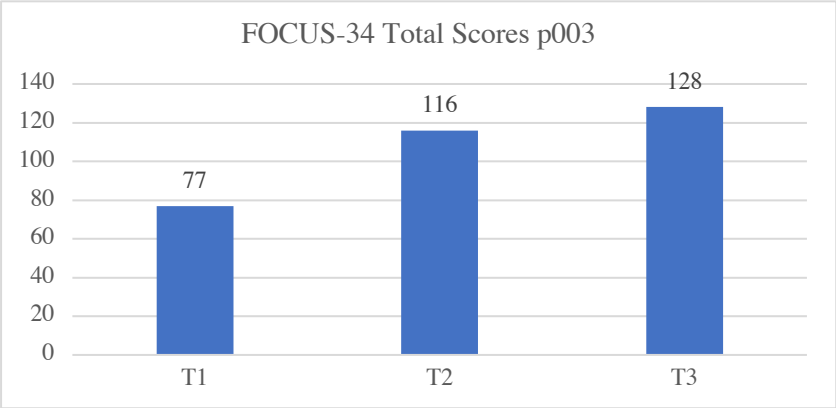


Figure 5: Participant 003 FOCUS-34



Participant 001: As can be seen in Figure 2, participant 001's ICS score increased by 1 point from T1 to T2, and again increased by 1 point from T2 to T3. Thus, there was no greater change following treatment than before treatment.

With respect to the FOCUS-34, for participant 001, the parent who completed the form did not complete one item each at T2 and T3 (item 1 at T2, and item 15 at T3). To ensure fair comparisons across time points, those two items were removed from all further analyses for all three timepoints. Participant 001's FOCUS score increased by 22 points from T1 to T2, and by 17 points from T2 to T3. According to the FOCUS-34 manual (Thomas-Stonell et al., 2012), a difference of 11 points or more is considered a clinically significant change. Thus, Participant 001 demonstrated clinically significant improvement across all three timepoints, although with no clear difference in magnitude of change before versus after treatment.

Participant 003: Participant 003 demonstrated no change from T1 to T2 on the ICS (total score = 25 at both timepoints). However, he did show an increase from T2 to T3 (T3 total score = 29).

Of note, Participant 003 was missing data from part 2 of the FOCUS-34 at T2. To be able to accurately analyze the data across the full time course, only part 1 of the FOCUS-34 was analyzed. The participant's FOCUS part 1 score increased by 39 points from T1 to T2, and by 12 points from T2 to T3. Thus, a clinically significant change was evident between each timepoint, although the greatest change occurred prior to onset of treatment.

Research Question 2: FOCUS-34 Capacity and Performance

To address Research Question 2 – whether the treatment had a differential impact on different domains of the WHO ICF model – the results of the FOCUS-34 were grouped by ICF domain and again analyzed visually and quantitatively. In cases where data were missing from any question or timepoint on the FOCUS-34, those data were eliminated across all timepoints to allow analysis across the full time course. This elimination of data inherently implies that some changes may not be reflected in the data, and scores are not directly comparable to those in the literature, but due to the large number of questions in each category, we are still able to determine if changes occurred within the larger categories of Body Function/Capacity and Performance. These data are presented in Figure 6 (p001) and 7 (p003).

Figure 6: FOCUS-34 Total Scores by WHO ICF Group for Participant 001

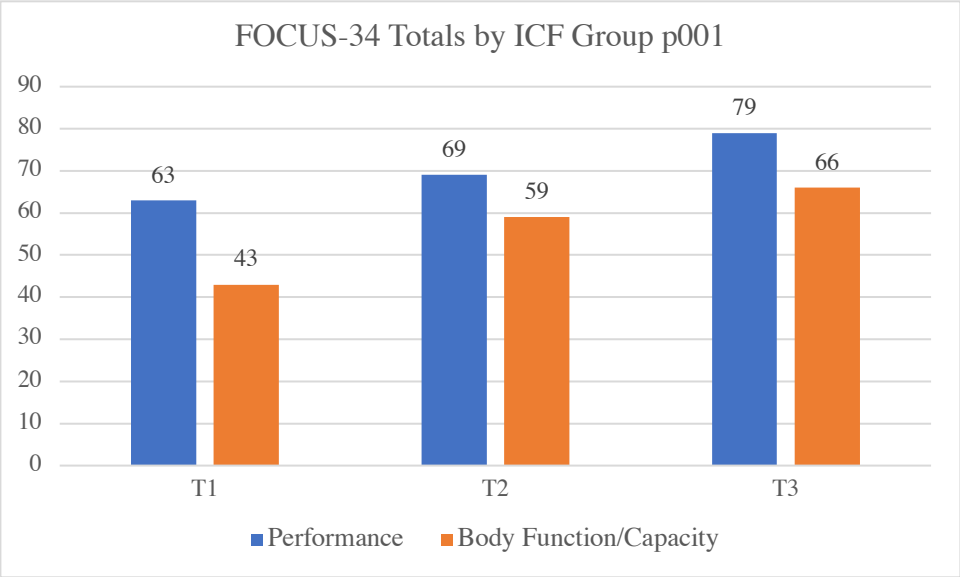
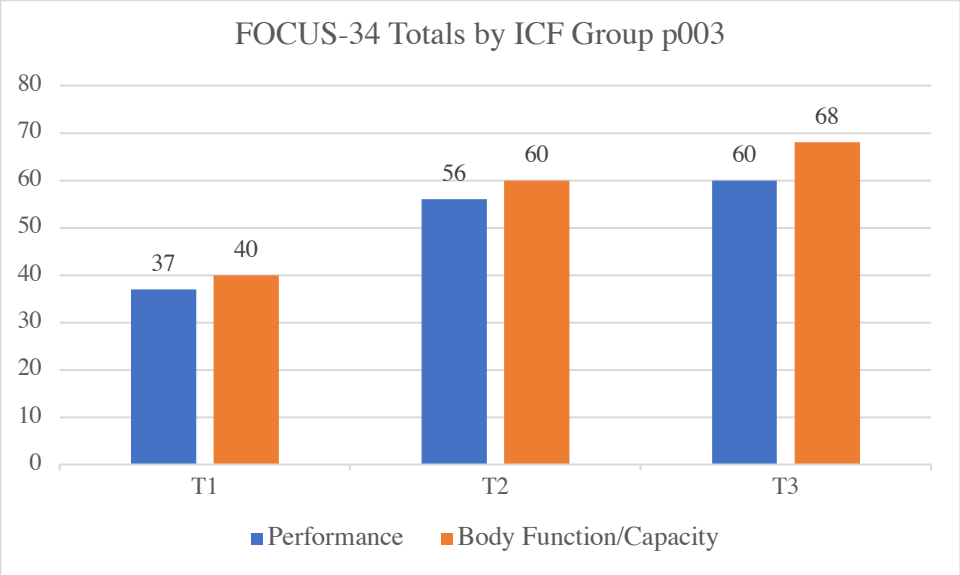


Figure 7: FOCUS-34 Total Scores by WHO ICF Group for Participant 003



Participant 001: As can be seen in Figure 6, the Body Function/Capacity total for Participant 001 increased by 16 points from T1 to T2, and by 7 points from T2 to T3. His Performance total increased by 6 points from T1 to T2, and by 10 points from T2 to T3.

Participant 003: Results for Participant 003 (see Figure 7) in the Body Function/Capacity domain showed an improvement of 20 points from T1 to T2, and an improvement of 8 points from T2 to T3. For the WHO ICF domain of Performance, Participant 003 was missing data at T2 that impacted all but one question in the social/play component of this domain. As previously stated, these data were eliminated across all timepoints to allow for consistency across all timepoints. With this taken into consideration, the data for Performance showed an improvement of 19 points from T1 to T2, and an increase of 4 points from T2 to T3.

CHAPTER 4

DISCUSSION

The purpose of this study was to examine potential changes in functional communication outcome measures as a function of Integral Stimulation treatment in two children with CAS. Specifically, we used the ICS (McLeod et al., 2012, 2015) and the FOCUS-34 (Thomas-Stonell et al., 2012), two parent-rated measures of communicative function and participation in everyday situations. The two children in this study had received approximately 16 hours of individual treatment on specific items that were selected to be personally meaningful. The primary data of this treatment study (speech accuracy) were presented elsewhere (Condoluci, 2020) and will not be discussed here in detail, except to contextualize the more generalized functional outcomes that are the focus of this study. First, we discuss findings from the overall scores (Research Question 1), followed by discussion of findings related to profiles in relation to the WHO ICF model of function (Research Question 2).

Generalized Functional Outcomes

The first research question looked at whether Integral Stimulation treatment results in generalized improvements in functional communication outcomes as judged by the ICS and FOCUS-34.

For Participant 001, there was no measured difference in ICS scores that can be attributed to the treatment. With respect to the ICS, only Participant 003 showed a larger improvement following treatment than before, whereas Participant 001 showed a small steady increase across timepoints. The pattern for Participant 003 is consistent with an effect of treatment, because the treatment was provided only between T2 and T3.

However, the increase was relatively small, and observed only in one of the two children, and therefore these results must be interpreted with caution and require replication.

The absence of a clear and consistent pattern indicating a generalized effect of treatment on the ICS does not necessarily imply that IS treatment did not, in fact, result in generalized improvements, but rather that there are additional factors to consider. First, the ICS tool may not have been sensitive to these improvements. Although the assessment has proven to be a valuable tool and its brevity is an advantage in administration, it is also possible that the question types and short length were not able to capture the potentially more subtle changes induced by treatment. Second, intervention was provided for a relatively short period of time, which may not be enough for observable changes outside of the context of treatment probes. Finally, Participant 001 demonstrated only relatively modest improvements in terms of speech accuracy (on average 10-20% increase across item sets), whereas Participant 003 did not show clear improvements in any treatment set (Condoluci, 2020). It is possible that if the children had shown greater and clearer improvements across a wider array of sets in treatment, improvements in these more generalized functional outcomes would be observed.

With respect to the FOCUS-34, both children demonstrated clinically significant improvements in each interval, but again, there was no indication that these improvements were due to the treatment. In fact, both children showed greater improvement from T1 to T2 (before treatment) than from T2 to T3 (after treatment). Thus, while the results overall are encouraging in that parents judged their children to show greater communicative participation, the reason for these improvements is not clear. Unlike the ICS, lack of sensitivity is unlikely in this case, since the FOCUS-34 has

more items and a wider scale (7-point scale vs. 5-point scale for ICS), and the FOCUS-34 clearly registered change. Thus, unlike the ICS, this assessment tool has greater sensitivity as its increased number of questions allows for a broader illustration of the child's functional communication skills. One possibility for the observed improvement is that the changes on the FOCUS-34 are related to maturation or other factors occurring in the children's life that are unrelated to the treatment. It is also possible that the reported change is due to parent bias in terms of anticipated improvement from being in a university-based research study. Such expectations could lead parents to rate their child's participation more favorably or to change their own behaviors in interacting with or observing their child. It is possible that the first completion of the FOCUS-34 sensitized parents to specific communicative participation behaviors and settings noted in the questions, and that this led them to pay closer attention to those behaviors and settings, and potentially to interact differently with their child as a result (e.g., encourage their use of words; encourage initiation of peer contacts).

It is important to note that in both the case of the ICS and FOCUS-34, the lack of clear improvement is not likely due to a ceiling effect. Participant 001 received an average score of 3.29 based on the 7-point Likert scale of the FOCUS-34. Similarly, he received an average score of 3.14 out of 5 points for the ICS. Participant 003 received an average score of 3.35 out of 7 on the FOCUS-34 and 3.57 out of 5 possible points on the ICS. Based on this information, there was ample opportunity and room for improvement to be reflected in the assessment tools and therefore there is little concern for a ceiling effect.

Either way, the improvement in overall FOCUS-34 score does not appear to be the result of the treatment. However, as previously discussed, this assessment was designed to encompass a variety of skills within communication and by solely looking at the total scores, these areas are not appropriately assessed. The results from this broad analysis further support the need for a more detailed analysis of communication outcomes based on the WHO ICF model which will be addressed in the discussion of Research Question 2.

WHO ICF Domain Outcomes

The second research question looked at whether the treatment has a differential impact on different WHO ICF domains (Performance and Body Function/Capacity), as captured by the FOCUS-34. For Participant 001, there was a measurable difference in the total score received on the FOCUS-34 for the Performance category of the WHO ICF model. This increase may be associated specifically with treatment because of the larger change noted from T2 to T3, during which treatment was administered. In the Body Function/Capacity domain, the participant demonstrated improvements across the three timepoints, however these cannot be attributed to treatment due to the smaller change noted following treatment as compared to the change prior to treatment.

For Participant 003, there is no measurable difference in the total scores received across either domain of the WHO ICF model that can be attributed to the treatment itself. Again, progress was noted across all three timepoints but due to the lack of notable change in scores between pre- and post-treatment, it cannot be concluded that treatment is the underlying cause of the progress.

In the case of Participant 003, it is important to note that all but one item was missing in the "social" subcategory of the Performance domain at T2. Again, to establish consistency, all of those items and any additional items that were missed at any timepoint were removed across all three timepoints. In total, this required the elimination of nine items from the Performance domain. Therefore, the changes observed in Participant 003 are inherently smaller. Additionally, it is possible that these nine questions addressed topics that were impacted by treatment but were unfortunately not reflected in the data.

Future Directions

The results of this study indicate that Integral Stimulation treatment may result in improvements within specific domains of the WHO ICF model. Although there may not have been observable changes in a broad analysis of generalization, treatment nonetheless impacted integral parts of children's communication success as indicated by the WHO ICF model. This finding indicates the need for future studies to analyze data in relation to more than just one area and investigate more functional implications. Future studies should replicate the analysis of data as it relates to the WHO ICF model to determine if additional participants demonstrate improvements in these functional communication outcomes.

Additionally, as previously stated, success in CAS treatment relies on frequent, intensive therapy over extended periods of time. Due to the relatively short nature of treatment in this study, future studies should investigate the effects of IS when administered for a longer period of time.

Finally, future studies should examine the relationship between accuracy on individualized speech targets and direct intelligibility as obtained from unfamiliar

listeners. Such studies should examine intelligibility of treated items as well as untreated utterances to determine how and to what extent gains on treatment targets impact intelligibility.

Conclusions

The present study examined changes in functional communication outcome measures based on parent report for two children with CAS who participated in a study of integral stimulation treatment. Both children showed some improvement in parent-reported intelligibility in context and in particular in communicative participation, over the course of the study period, indicating that gains in more generalized functional outcomes are possible and measurable in children with CAS even over a relatively short period of time.

However, the change in parent-rated intelligibility could reasonably be attributed to treatment only for one child (p003). Similarly, improvements in communicative participation could not be confidently related to the treatment, because gains after treatment were no greater than those before treatment. Inspection of subscores related to different WHO ICF domains did not reveal consistent patterns; for one child (p001), the Performance items appeared to show a greater improvement after treatment than before treatment, whereas for the other child (p003) both the Capacity domain and the Performance domain showed greater improvement before treatment. It should be noted that these data may underestimate or miss effects, given that a considerable portion of the FOCUS-34 was not completed for Participant 003. Further research with larger sample sizes is needed to examine the relationships between outcome measures and understand the functional impact of integral stimulation treatment for CAS.

One final important conclusion from this study is that gains in speech accuracy on individualized probes do not necessarily predict changes in more functional outcome measures: Participant 001 made clear gains on accuracy in most of his individualized target utterances yet showed little to no clear treatment-related improvement on these parent-reported measures. In contrast, Participant 003 did not show reliable changes in speech accuracy yet showed considerable improvements in communicative participation and an improvement in parent-rated intelligibility that was greater following treatment than before treatment. In other words, the findings suggest that measures at different levels of the WHO ICF model dissociate to some extent, and that we cannot assume that gains in speech accuracy will be associated with improved functional communication.

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APPENDIX

ICS and FOCUS-34 Scores

		Participant 001	Participant 003
ICS Total Score	T1	22	25
	T2	23	25
	T3	24	29
FOCUS-34 Total Score	T1	106	77
	T2	128	116
	T3	145	128
FOCUS-34 *Performance sub score*	T1	63	37
	T2	69	56
	T3	79	60
FOCUS-34 *Body Function/Capacity sub score*	T1	43	40
	T2	59	60
	T3	66	68