Self-Reported Communication Attitudes of Children with Childhood Apraxia of Speech

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ABSTRACT

Much of the research literature on childhood apraxia of speech (CAS) has focused on understanding, diagnosing, and treating the impairment, rather than examining its broader impact. The present study focuses on the Personal Factors component of the World Health Organization model. Two validated communication attitude questionnaires were administered to 12 children with CAS enrolled in an intensive speech-focused intervention. Children's scores were compared to the questionnaires' typically developing norms. Relationships to CAS severity, caregiver perceptions of communicative participation, frustration ratings during therapy, and change over a brief period were also investigated. Preliminary findings indicate that older but not younger children with CAS are more likely to have greater negative self-perceptions about their speech. No significant correlation was found between caregivers' perceptions of communicative participation in various contexts and communication attitudes, highlighting the need to include more child self-report measures in research. Further implications for CAS assessment and intervention are discussed.

INTRODUCTION

Capturing child perspectives can provide researchers and clinicians with a unique understanding of the contextual factors that influence how children experience the world. This is particularly important for children with speech sound disorders who have distinct communicative challenges compared to their typically developing peers. Speech sound disorders (SSD) are a group of disorders that cause difficulty with motor production, perception, or phonological representations of speech sounds or syllables (American Speech-Language-Hearing Association, [ASHA] 2007). Examples of SSDs include articulation disorder, phonological disorder, and childhood apraxia of speech. Articulation disorder impacts speech production at the sound level, while phonological disorder impairs production at the syllable level (Dodd, 2014). These disruptions cause predictable errors like sound substitutions and consonant cluster reductions.

Childhood apraxia of speech (CAS), a motor-based speech sound disorder, results from inaccurate motor planning and programming of the speech musculature (ASHA, 2007). Though estimates of prevalence are ongoing, approximately 1-2 children per 1,000 in the United States are thought to be diagnosed with CAS (Shriberg et al., 2019). Children with CAS have difficulty timing and orienting their speech musculature that cannot be accounted for by physical or cognitive disabilities (Murray & Iuzzini-Seigel, 2017). The inability to accurately plan movements causes several impairments including prosodic and stress errors, inconsistency over repeated trials, and consonant and vowel distortions (Lewis et al., 2004). The unpredictability of impairments poses a distinct challenge for treatment and distinguishes a CAS diagnosis from other speech sound disorders. Lewis and colleagues (2004) found that school-age children with CAS have more frequent and severe errors compared to their peers with other SSDs. A CAS diagnosis also puts children at an increased risk of other expressive and receptive difficulties

including reading and writing (Lewis et al., 2004). These distinguishing factors may mean that CAS's impact on children's daily lives and personal attitudes differ from the experience of children with other SSDs.

The World Health Organization's International Classification of Functioning, Disability, and Health (ICF) provides a framework for better understanding the complex array of factors that impact individuals with various disorders (see *Figure 1*, Threats, 2006). This framework recognizes the interconnections between the three levels and encourages clinicians to approach a client's care holistically. Level (1) is the identification of the disorder or disease. Level (2) consists of body functions/structures, activity, and participation. CAS research thus far has almost exclusively addressed Level (2) of the ICF, with many studies aimed at finding effective treatments. Though no treatment has conclusive evidence of effectiveness, some show promising outcomes. Current evidence suggests that the best CAS treatments target motor learning principles to improve overall accuracy across syllables and words (Murray & Iuzzini-Seigel, 2017). Effective CAS treatments should also be deliberate in both how children practice during therapy and what targets are chosen (Maas et al., 2014). One such treatment is Dynamic Temporal and Tactile Cueing (DTTC), which relies on Integral Stimulation to target the planning and programming of multiple speech sounds. A key feature of DTTC is its use of individualized meaningful target words (Strand, 2020). In contrast, Rapid Syllable Transition treatment (ReST) consists of varying the stress of nonsense words and probing for real words (McCabe et al., 2017). The rationale for ReST is that nonsense targets can lead to more accurate planning across multiple generalizable words. Outcomes measures for both DTTC and ReST are target accuracy. In addition to target accuracy, other factors such as intelligibility, activity and participation, and

changes in personal attitudes towards communication should also be considered to determine

degree of treatment success.

Figure 1*: The World Health Organization's International Classification of Functioning, Disability, and Health*
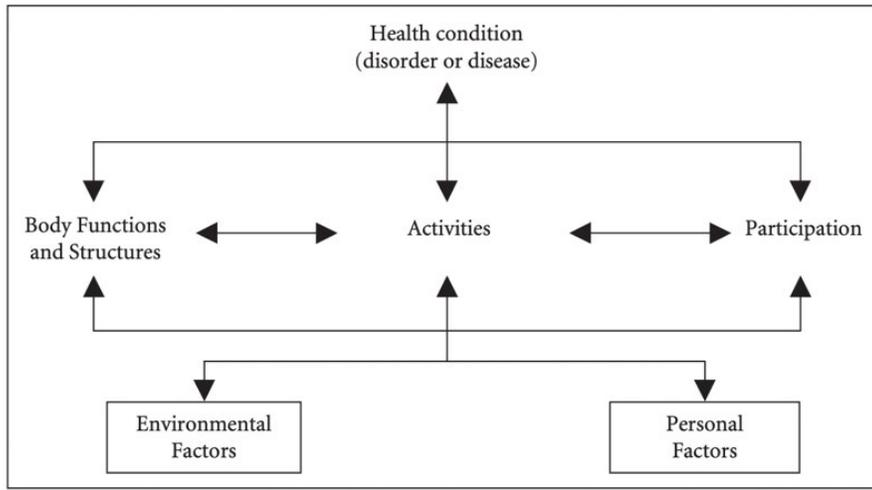
Neglected from current research on CAS is Level (3) of the ICF: environmental and

personal factors. Environmental factors are described as those influences that play a significant

role in the individual's life beyond personal control. For instance, external factors such as public

policy or parental support can impact an individual's functioning. Personal factors are those

more unique to the person's life such as race, ethnicity, personality, and motivations (Threats,

2006). They can be divided into unchangeable factors (e.g., race and age) and factors that have

the possibility of changing such as lifestyle habits and coping styles (Howe, 2008). Personal

factors can help researchers and clinicians understand how different individuals are impacted by

the same diagnosis (Threats, 2006). Understanding the perspectives of children with CAS may

prove useful for informing treatment approaches and for developing functionally relevant goals

(Murray & Iuzzini-Seigel, 2017). Given that personal factors were found to influence treatment outcomes for children with phonological disorder (Baker & McLeod, 2011a, 2011b), it is critical that researchers better understand how this translates for children with CAS.

Parent perspectives are often used as a proxy for understanding the feelings and thoughts of children with communication disorders. This is usually due to the perceived and inherent difficulties that accompany collecting child perspectives. These difficulties include questioning the reliability and validity of a child's response and accounting for extra ethical considerations when working with children (Harcourt, 2011). In many cases, parents provide accurate and detailed accounts for their child. For example, Rusiewicz et al. (2018) used parent measures to evaluate the daily impact of a CAS diagnosis, with parents being particularly concerned about their child's overall intelligibility and ability to navigate social situations. More generally, McCormack et al. (2010) asked parents and speech-language pathologists to rate how they felt about their children's speech impairment using an ICF-CY based questionnaire. They found that both populations expressed the greatest concern regarding interpersonal relationships and verbal communication skills. These studies demonstrate the ability of parent perspectives to collect more detailed information than would usually be possible from a child.

Solely collecting parent perspectives, however, relies on the assumption that they are always accurate. Investigations into correlations between parent and child perspectives for children with phonological disorder (McCormack et al., 2019) and children who stutter (Vanryckeghem, 1995) have shown that parents do not always understand their child's communication attitudes. Disregarding child perspectives also fails to directly include children in the conversation. According to Article 12 of the United Nations Conventions on the Rights of the Child (1989) and the follow-up General Comment No. 7 (2005), children must be allowed to

express their views and be included in decisions that directly impact them (Lundy et al., 2011).

These declarations underscore and reinforce the need to incorporate more child perspectives into

research and clinical practice in communication disorders. When collected correctly, children

can provide unique, distinct insights into their life experiences (Harcourt, 2011). It is worthwhile

to acknowledge that children's perceptions may change frequently and reflect growing

development, but this does not imply that their responses at the given assessment are any less

relevant or valuable to examine (Harcourt & Einarsdóttir, 2011). Directly asking children with

speech and language disorders how they feel is also an effective way of understanding if children

are receiving adequate support (Roulstone & McLeod, 2011).

Relatively few self-report measures exist for understanding the self-perceptions of

children with communication disorders. Each have distinct strengths and weaknesses. The lack

of available measures is a contributing factor as to why there are few studies in this realm. For

the current study, relevant measures were examined for validity and feasibility.

The Speech Participation and Activity Assessment of Children

The Speech Participation and Activity Assessment of Children (SPAA-C) provides open

ended and Likert scale questions to uncover how speech difficulties influence children's lives

(McLeod, 2004). It considers how other members of the child's life (i.e., parents, friends,

siblings, parents, teachers, others) perceive the child with the speech difficulty. Questions range

from "Who do you like to play with?" to "Do you think your talking is different from other

children's?" to gain a larger picture of the child's perceptions. It is designed to be adapted for the

particular needs of the researcher and abilities and age of the child. McLeod, Daniel, and Barr

(2013) used the SPAA-C with school-aged children with speech sound disorders and emphasized

that children would be better supported if their perspectives were more often incorporated in

assessment and intervention. Other applications of the SPAA-C have revealed the frustrations of the child with an SSD in being misunderstood and communicative partners lack of listening skills (McCormack, McLeod, Harrison et al., 2010) and incapacity to provide adequate support in various settings (Daniel & McLeod, 2017). Though valuable, application of the SPAA-C can be burdensome and time-consuming for participants and is less optimal for quantitative analysis.

The Pictorial Scale of Perceived Competence and Social Acceptance for Young Children

The Pictorial Scale of Perceived Competence and Social Acceptance for Young Children (PSPCSA) asks children to compare pictures, a useful tool when examining child perspectives (Elden, 2013), and decide which picture is more representative of themselves (Harter & Pike, 1984). A score of 1 to 4 is given for each image depending on the extent the child indicates that picture applies to them. The PSPSCA has been given to children with specific language impairment (SLI), with findings suggesting that older children with SLI struggle more with scholastic competence, social acceptance, and behavioral conduct than their typically developing peers (Jerome et al., 2002). However, question wording on the PSPSCA is outdated (i.e., "maternal acceptance" rather than "parental acceptance"), and there are doubts regarding its application to diverse populations (Fantuzzo, 1996).

The Communication Attitude Test for School-age Children who Stutter

The Communication Attitude Test for School-age children who stutter (CAT), a component of the Behavior Assessment Battery for Stuttering, is designed to assess the speech attitudes of children ages 6;0 and older (Brutten & Dunham, 1989; Brutten & Vanryckeghem, 2007). It consists of 33 true/false statements such as "I like to talk" and "My parents like how I talk." Jones and colleagues (2021) found the CAT to possess among the highest measurement properties compared to other measures of the psychological impacts of stuttering.

The Communication Attitude Test for Preschool Age Children who Stutter

The Communication Attitude Test for Preschool Age children who stutter (KiddyCAT) was developed as an extension of the CAT to assess how preschool children feel about their speech (Vanryckeghem & Brutten, 2007). It asks children (ages 3;0-5;11) 12 yes/no questions such as "Do you like how you talk?" and "Do words sometimes get stuck in your mouth?"

Both the CAT and KiddyCAT have been administered in several countries including Australia (McCormack, McLeod, & Crowe, 2019), Italy (Bernadini et al., 2009), Belgium (Vanryckeghem & Brutten, 1992; Vanryckeghem, De Niels et al., 2015), Japan (Kawai, 2012), and Sweden (Johannisson, 2009), implicating its validity across cultures. Though developed for stuttering, these assessments have been successfully administered to many clinical populations including to children with phonological disorder (McCormack, McLeod, & Crowe, 2019) articulation disorder (Luc & Brutten, 1990) and cleft palates (Havstam et al., 2011). These applications are positive indications that the CAT and KiddyCAT can be administered to populations other than to children who stutter.

Administration of the CAT and KiddyCAT to children who do and do not stutter have revealed that children who stutter are more likely to have negative speech perceptions and that younger children tend to feel more positively than older children about their speech (Brutten & Vanryckeghem, 2007). Influence of age differences beyond stuttering was highlighted by McCormack, McLeod, and Crowe (2019), who found that preschool age children with phonological disorder did not have more negative perceptions of their speech compared to the established norms.

Those administering both assessments are provided specific directions, which minimizes issues with test-retest reliability and distortion of results. The CAT and KiddyCAT were chosen

for the present study because they are standardized, encompass the age range (4;0-10;0), are easy to track changes over time, and have been found to be reliable and valid across a variety of languages, cultures, and clinical populations. Despite its widespread use, neither the CAT nor KiddyCAT have been administered to a group of children with CAS. Given that CAS differs in consistency of impairment and response to treatment compared to other SSDs, it is clearly necessary to further understand these children's communication attitudes.

The Present Study

The present study was designed to fill this critical gap in the literature by administering the CAT and KiddyCAT to children with CAS. This study took place in the context of a randomized controlled trial of (impairment-focused) speech therapy for CAS, which was conducted as an intensive summer camp (see Methods section for further description of the broader study context). This setting afforded the opportunity to address the following questions:

1. Do communication attitudes of children with CAS differ from those of typically developing children, based on the tests' normative values?

2. Do communication attitudes of children with CAS relate to impairment severity, as operationalized by DEMSS score?

3. Do communication attitudes of children with CAS relate to caregivers' perceptions of their child's communicative participation, as operationalized by FOCUS-34 score?

4. Do communication attitudes of children with CAS relate to average frustration level during therapy?

5. Do communication attitudes of children with CAS change over the course of a brief intervention period?

METHODS

Study Context

This study took place in the context of a larger ("parent") study involving a randomized controlled trial to test initial efficacy of ASSIST (Apraxia of Speech Systematic Integral Stimulation Treatment; Maas et al., 2019) for children with CAS. ASSIST is an impairment-focused speech treatment approach based on integral stimulation ("watch me, listen to me, say what I say") and principles of motor learning (Maas et al., 2008). Speech targets are words and phrases selected based on personal functional relevance and contain only sounds in the child's phonetic inventory. The treatment involves drill-based repetition of targets, with feedback and cues provided by the treating clinician to help the child achieve movement patterns for accurate speech.

In this parent study, children participated in an intensive 4-week virtual summer camp program, during which they received 16 hours of individual ASSIST in 32 sessions of 30 minutes each. Children were randomly assigned to receive all their individual treatment in the first two weeks (immediate massed), the second two weeks (delayed massed), or divided over all four weeks (distributed) (see *Figure 1* for an overview of the design of the parent study). Group activities and individual ASSIST were delivered via videoconference. Before, at midpoint, and after the 4-week period, children attended individual data collection sessions in person in the Speech, Language, and Brain Lab at Temple University. Details and results from the parent clinical trial are not further reported here, except insofar as relevant to the present study on child-reported outcome measures (CAT and KiddyCAT), which were administered at T2 and T3. All study procedures were approved by the Temple University IRB (protocol #25807) and informed consent was obtained from at least one parent and all children provided assent.

Figure 1. Design of the clinical trial that forms the context for the present study. Data collection (CAT and KiddyCAT) occurred in Week 4 (T2) and Week 7 (T3).

Group	Condition	wk1	wk2	wk3	wk4	wk5	wk6	wk7
Massed ASSIST (N=6)	Immediate (N=3)	T1	8 hrs	8 hrs	T2			T3
	Delayed (N=3)					8 hrs	8 hrs	
Distributed ASSIST (N=6)	Distrib. (N=6)		4 hrs	4hrs		4 hrs	4 hrs	

Participants

Twelve children ages 4-10 years old with a primary diagnosis of CAS participated in this study. Three expert speech-language pathologists (SLPs) provided independent ratings of presence of CAS, using a 3-point scale where 0 = no CAS, 1 = possible CAS, and 2 = CAS. These ratings were based on the presence of current consensus features of CAS (ASHA, 2007), including inconsistent errors on consonants and vowels, difficulties achieving or transitioning into articulatory configurations, and abnormal prosody such as lexical stress errors. At least one SLP made this judgment on the basis of the live assessment sessions, whereas the remaining SLPs made their judgments from video-recordings of the assessment sessions. To be included in the study, children were required to receive an average rating of >1 across the three expert SLPs (M=1.69) and an Apraxia score of 1 or 2 on a Maximum Performance protocol (Thoonen et al., 1996, 1999), the only prospectively validated protocol to date with acceptable sensitivity and specificity.

Inclusion criteria were (a) English as primary home language per parent report, (b) normal hearing as determined by parent report and/or pure tone audiometric screening, (c) nonverbal cognition in the typical range based on the Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003), and (d) verbal output of 50+ words and communicative intent, per SLP and parent report. Exclusionary criteria were (a) co-occurring neurobehavioral diagnosis (e.g., autism) per parent report, (b) significant visual impairment per parent report, (c) primary

speech diagnosis of dysarthria based on SLP judgment, (d) structural-anatomical abnormalities of the oral structures (e.g., cleft palate) based on parent report and SLP judgment from an oral mechanism evaluation, and (e) inability to operate or attend to virtual sessions, per parent and SLP judgment.

A battery of tests were administered prior to allocation to groups to determine eligibility and to characterize participant profiles. These are reported in ***Table 1***, but most of these will not be discussed in detail here, except for the Dynamic Evaluation of Motor Speech Skill (DEMSS; Strand & McCauley, 2019), as the DEMSS score represents an index of CAS severity that will be used in the subsequent analyses. The DEMSS is a criterion-referenced tool designed to assess children, ages 3 and older, with moderate to severe speech impairment, prosodic or vowel errors, poor speech intelligibility, or who have minimal or no verbal communication skills (Strand & McCauley, 2019). The assessment can confirm or rule out a CAS diagnosis, be an estimate of severity and prognosis, and/or demonstrate the effectiveness of various cues. Administration generally takes less than 30 minutes. The test requires the child to imitate an SLP across 60 utterances. The child is given additional support and cues if the initial attempt is incorrect. Possible scores range from 0 to 426, where lower scores indicate greater severity. Scores between 0 and 323 indicate significant evidence for CAS, scores from 324 to 373 indicate some evidence of at least mild CAS, and scores from 374 to 426 reflect little to no evidence for CAS (Strand & McCauley, 2019).

Participants were recruited through local community referrals, existing databases of individuals who expressed prior interest in research opportunities, community outreach and education events, and advertisements in newsletters and websites. Of the 16 children assessed for

eligibility, 12 children met the selection criteria, including 9 boys and 3 girls (see *Figure 1* and

Table 1).

Table 1. Participant information.

Child	Age[a]	Sex	Cond.[b]	CAS rating[c]	DEMSS score[d]	GFTA[e] raw	GFTA[e] SS	DEAP[f] Phon. SS	DEAP[f] Incons.	RIAS NIX[g]	RIAS NV Memory[h]	EVT[i] SS	PPVT[j] SS	CELF[k] CLS
301	6;0	M	D	1.00	383	43	63	60	32%	50	50	82	96	75
306	4;2	M	D	2.00	215	107	49	65	80%	83	63	117	93	NA[l]
309	7;10	M	D	1.33	298	73	40	55	68%	42	64	81	85	66
310	4;1	M	D	2.00	DNC[m]	131	40	CNC[n]	72%	66	75	83	71	NA[l]
314	10;0	M	D	2.00	401	22	59	NA[l]	40%	40	38	80	95	72
322	8;3	F	D	1.00	395	10	70	105	36%	42	61	87	86	93
304	5;5	M	M1	2.00	295	40	72	75	44%	78	56	98	90	93
312	5;9	F	M1	1.67	143	109	40	55	64%	47	81	81	76	79
321	6;4	M	M1	2.00	262	83	40	55	56%	43	49	89	94	93
305	7;0	F	M2	2.00	382	70	40	55	44%	46	49	86	78	75
313	4;11	M	M2	2.00	DNC[m]	78	49	65	84%	67	59	96	86	NA[l]
324	5;2	M	M2	1.33	357	76	47	65	52%	53	66	98	118	100

[a] Age at T2 in years; months
[b] Condition: D = Distributed; M1 = Massed Immediate (phase 1); M2 = Massed Delayed (phase 2)
[c] Mean of 3 expert speech-language pathologists' independent ratings regarding presence of CAS (0 = no CAS; 1 = possible CAS; 2 = CAS)
[d] Score on the Dynamic Evaluation of Motor Speech Skill (Strand & McCauley, 2019)
[e] Raw score and Standard Score (SS) on the Goldman-Fristoe Test of Articulation, 3rd Edition (Goldman & Fristoe, 2015)
[f] Diagnostic Evaluation of Articulation and Phonology (Dodd et al., 2006) phonology standard score and inconsistency percentage.
[g] Nonverbal Intelligence Quotient (NIX) T-score based on the Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003)
[h] Nonverbal Memory T-score based on the Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003)

[i] Standard score (SS) on the Expressive Vocabulary Test, 3rd Edition (Williams, 2019)
[j] Standard score (SS) on the Peabody Picture Vocabulary Test, 5th Edition (Dunn, 2019)
[k] Core Language Score (CLS) Standard Score on the Clinical Evaluation of Language Fundamentals, 5th Edition (Wiig et al., 2013)
[l] NA = not administered due to age outside normative range
[m] DNC = did not complete (no total score available)
[n] CNC = could not compute (score outside of normative range)

Instruments

CAT and KiddyCAT

As described in the Introduction, the CAT and KiddyCAT have been used to provide insight into how children with a range of speech disorders feel about their speech. The CAT is designed for children 6 and older and consists of 33 true/false statements to be answered by the child. The KiddyCAT is designed for children between 3 and 6 years old and consists of 12 yes/no questions to be answered by the child. Both the CAT and KiddyCAT include statements/questions where an approximately even number of true and false (or yes and no) responses indicate negative self-perceptions. Each answer that indicates a negative self-perception is awarded one point. Thus, higher scores on both assessments indicate more negative speech perceptions. The average KiddyCAT score for children who do not stutter is 1.79 (SD=1.78) and the average CAT score for children who do not stutter is 6.38 (SD=5.21). Per each manual, a score of 5 or higher on the KiddyCAT or a score of 17 or higher on the CAT is indicative of a negative speech perception. However, clinicians and researchers are also advised to consider and investigate slightly lower scores on both assessments during analyses.

Focus on Outcomes of Communication Under Six (FOCUS)-34

The Focus on Outcomes of Communication Under Six (FOCUS-34; Oddson et al., 2019; Washington et al., 2012) is a parent-rated 34-question assessment of child participation across various communicative contexts. It consists of two parts: Part 1 asks about how well a statement applies to the child, and Part 2 asks about the amount of help a child needs to accomplish communicative tasks. Example items from Part 1 include "My child is confident communicating with adults who do not know my child well" and "My child uses words to ask for things". Each

item is scored from 1 (not at all like my child) to 7 (exactly like my child). Example items from Part 2 include "My child is included in games by other children" and "My child joins in conversations with other children". Items are rated from 1 (cannot do at all) to 7 (can always do without help). The FOCUS-34 takes approximately ten minutes to complete.

All participants' parents in the current study, regardless of age, were given this form at each time point. While this assessment was designed and normed for children under age six, there is no significant reason to question its validity for older children, and there is precedent for use of the FOCUS-34 with children older than six (e.g., Rusiewicz et al., 2018). The same parent completed the form at each time point.

Frustration Ratings

As part of the parent study's safety protocol, potential negative side effects of treatment were monitored, with a particular emphasis on child frustration. The treating clinician provided a numerical score reflecting the child's frustration level, based on their judgment informed by their knowledge of the child and the child's behavior in the session. Because frustration may be expressed differently by different children (e.g., some children may cry, others may refuse to continue, others may display avoidance behaviors), judgment by the treating clinician was deemed to be an adequate measure to capture these potentially different expressions. Possible scores were 0 (no frustration, full compliance with procedures throughout the session), 1 (some frustration, occasional noncompliance during session), 2 (significant frustration, frequent noncompliance during session), and 3 (marked frustration, noncompliance during entire session).

<u>Procedures</u>

At the end of T2 and T3 data collection sessions, trained research assistants, blinded to treatment status, administered the CAT (for children 6;0 and older; n=6) or KiddyCAT (for children 4;0-5;11; n=6). Per the CAT and KiddyCAT manual, children were instructed that there were no right or wrong answers because the questions were about what they thought about their own speech. Before administration, two practice questions were given to establish adequate understanding of the task. Prior to T2 administration, any child given the KiddyCAT and deemed to have a questionable understanding of the concepts 'easy' and 'difficult' was provided two jars and asked to distinguish which was easy to open and which was hard to open (n=1). The child had no trouble understanding this concept.

Each question was read aloud by research assistants for both the CAT and KiddyCAT. Participants were required to respond true/false for the CAT and yes/no for the KiddyCAT. For the CAT, right/wrong and yes/no were also deemed acceptable responses in place of true/false. Administration took approximately 5-15 minutes for the KiddyCAT and 15-30 minutes for the CAT. All sessions were audio- and video-recorded for subsequent analysis and reliability check.

<u>Reliability</u>

CAT and KiddyCAT questionnaires were scored independently by a second scorer (who was not blinded to condition) from session recordings. Each answer was screened for adequate understanding. A few specific questions were excluded due to missing items; for this reason, all scores were converted to percentages for analysis.[1] The scores of the second scorer were

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[1] At T2, two CAT questions were excluded for Participant 309, one CAT question was excluded for Participant 322, and one KiddyCAT question was excluded for Participant 310. At T3, one CAT question was excluded for Participant 309 and Participant 314. All analyses were conducted both on raw scores and percentaged scores; the pattern of results did not differ in any analysis.

compared with the original forms scored by the examiner who administered the questionnaires[2] to evaluate inter-rater reliability. Inter-rater reliability of total scores was assessed via intraclass correlation coefficients (ICC) with 95% confidence intervals, calculated with the *irr* package (Gamer et al., 2012) in R software (R Studio version 1.2.5042), based on a single-measures, absolute agreement, two-way mixed effects model (McGraw & Wong, 1996a, 1996b). Resulting ICCs were in the good to excellent range (Koo & Li, 2016) for all four comparisons (CAT T2, CAT T3, KiddyCAT T2, KiddyCAT T3; see *Table 2* below).

Discrepancies between scorers were reviewed and resolved by a third independent scorer and were attributed to human error by the administrator. For all subsequent analyses, scores from the second scorer were used because this scorer had opportunity to re-watch the video to increase confidence in the child's response.

Table 2. Interrater reliability across tests and timepoints based on total score converted to percentage. ICC = intraclass correlation coefficient; CI = confidence interval [lower bound, upper bound]. Interpretation of ICC values based on Koo and Li (2016).

Test	T2			T3		
	ICC	*95% CI*	*Interpretation*	*ICC*	*95% CI*	*Interpretation*
KiddyCAT	0.836	[0.304, 0.975]	good	0.886	[0.339, 0.987]	good
CAT	0.896	[0.397, 0.985]	good	0.995	[0.962, 0.999]	excellent

<u>*Analyses*</u>

The research questions were addressed as follows. For all inferential statistical analyses, an alpha level of 0.05 was adopted to determine statistical significance.

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[2] During T2, the 35-question form of the CAT was administered to 3 participants. The updated 33- question version was administered to all participants at T3. The additional two questions from the 35-question form were not considered in analyses. The KiddyCAT form was consistent across both timepoints.

Research Question 1 (comparison with normative values) was addressed descriptively. Specifically, individual CAT and KiddyCAT total scores (percentages) at each timepoint as well as their means across the two timepoints were compared to their respective normative values (also converted to percentages for this comparison), and children whose scores were more than 1 standard deviation from the normative mean were identified.

For Research Question 2 (relation with CAS severity), CAT and KiddyCAT scores at each timepoint and their means across timepoints were correlated with the DEMSS total score using non-parametric Spearman correlations, given the small sample sizes. Note that more severe CAS is reflected by *lower* DEMSS scores. Thus, if more severe CAS is associated with more negative self-perceptions, we expect a negative correlation, because more negative self-perceptions are reflected by *higher* CAT/KiddyCAT scores.

For Research Question 3 (relation with parent perception of communicative participation), CAT and KiddyCAT scores at each timepoint were correlated with the FOCUS-34 total scores from those same timepoints using non-parametric Spearman correlations. If children with more negative self-perceptions are less likely to participate in communicative activities (as judged by their parent), then we expect a negative correlation, because higher FOCUS-34 scores indicate greater communicative participation.

For Research Question 4 (relation with frustration during treatment), CAT and KiddyCAT scores at each timepoint as well as their means across timepoints were correlated with the mean frustration rating across all treatment sessions and with the percentage of sessions during which children received a frustration rating of 3 (marked frustration). If children with more negative self-perceptions are more prone to frustration, we expect a positive correlation.

For Research Question 5 (change following brief intervention period), descriptive analysis was used given the small and unequal sample sizes in the different treatment conditions. The number of children whose scores increased, remained the same, and decreased numerically were identified.

RESULTS

Results are described below by research question. Within each research question, CAT and KiddyCAT results are described separately. For the KiddyCAT, one participant (P306) did not return for T3 testing; thus, analyses involving T3 are based on data from the remaining five children in this age range.

Research Question 1: Comparison to Normative Values

Figure 2: KiddyCAT Scores
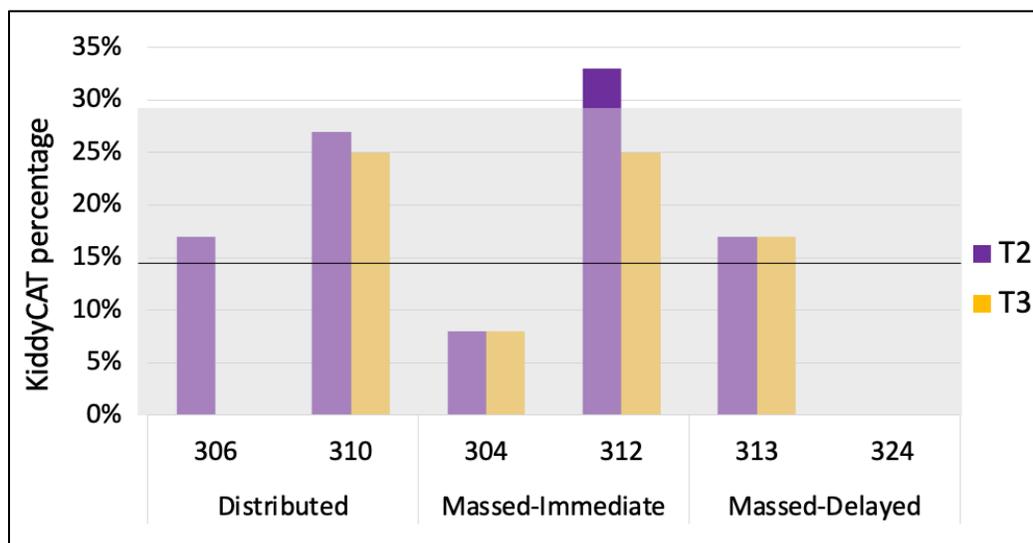
Figure 3: CAT Scores
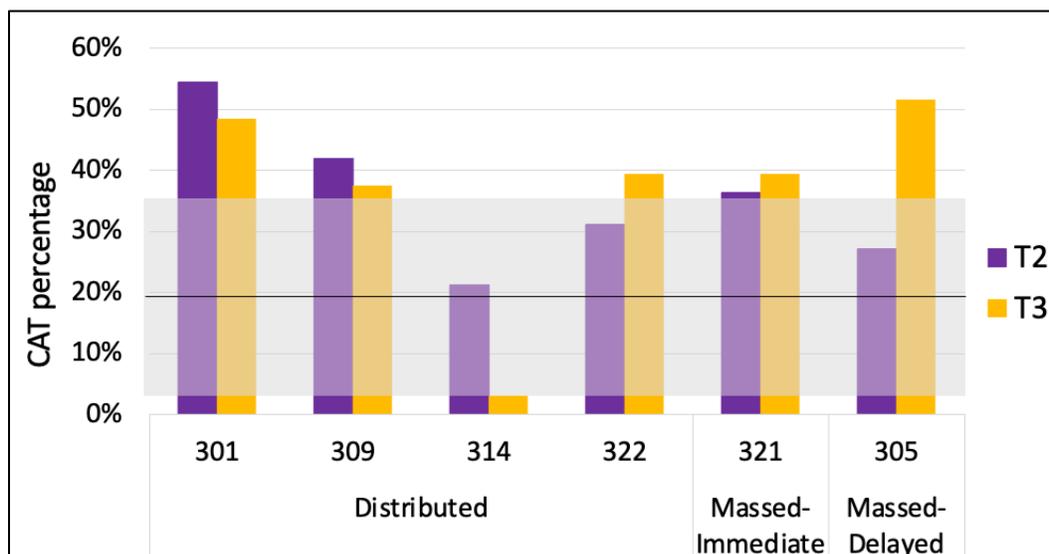
Among the younger children (KiddyCAT), one child fell above the normative range at T2, but none did at T3. One child (P324) fell below the normative range at both timepoints.[3] Among the older children (CAT), 3 out of 6 children had higher (i.e., more negative) scores compared to the normative sample at T2, and 5 out of 6 children at T3; one child showed a lower (i.e., more positive) score at T3 compared to the normative sample.

Per the CAT and KiddyCAT manual, participants' scores on these assessments were also compared to +/- 2 standard deviations from the normative range. For the KiddyCAT, no child fell outside of 2 standard deviations of the mean (a score of 5 or above) at either timepoint. For the CAT, one participant at T2 (P301) and one participant at T3 (P305) fell above 2 standard deviations of the normative range (a score of 17 or above).

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[3] P324 scored 0 at both timepoints, whereas P306, who did not return for testing, does not have a score at T3.

Research Question 2: Relationship with CAS Severity

Figure 4: Comparison to DEMSS Scores
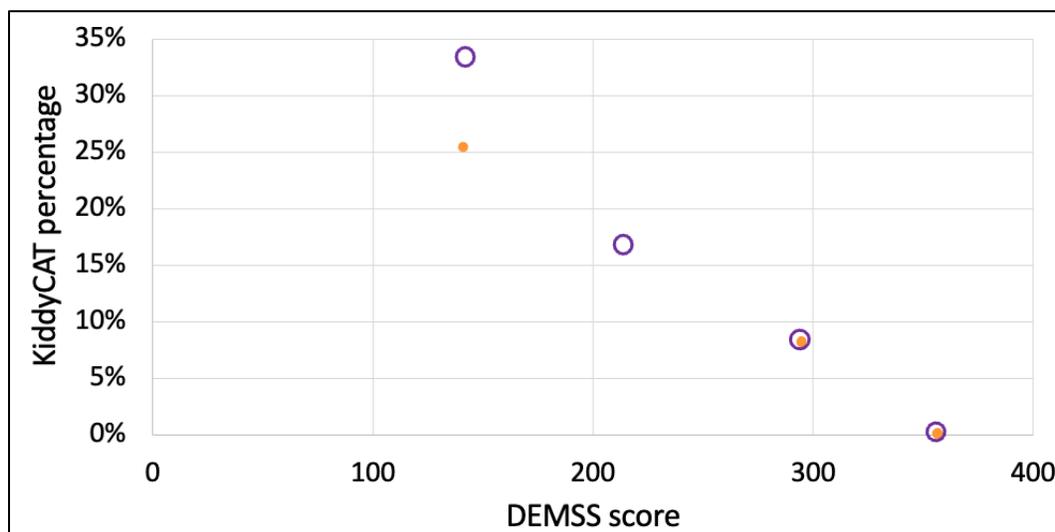
KiddyCAT% x DEMSS: T2 & T3

For the CAT, correlations with baseline DEMSS scores were negative but none were significant (T2: rho = -0.486, p = 0.329; T3: rho = -0.261, p = 0.618; mean: rho = -0.486, p = 0.329). For the KiddyCAT, correlations with baseline DEMSS scores were negative and also significant (rho = -1.000, p < 0.0001, for T2, T3, and mean). It should be noted that these correlations are based on small sample sizes (n = 4 for T2, n = 3 for T3 and mean) due to missing T3 data for Child 306 and incomplete DEMSS scores for two other children.

Research Question 3: Relationship with Parent-Rated Communicative Participation

For the CAT, a moderate negative correlation was observed at T2, but this correlation was not significant (rho = -0.657, p = 0.156). At T3, a small and nonsignificant negative correlation was observed (rho = -0.232, p = 0.658).

For the KiddyCAT, analysis revealed a small but nonsignificant positive correlation at T2 (rho = 0.176, p = 0.738) and a small nonsignificant negative correlation at T3 (rho = -0.103, p = 0.870).

Research Question 4: Relationship with Frustration Level during Treatment

For the CAT, small to moderate positive correlations were obtained with mean frustration scores; however, none of these correlations were significant (T2: rho = 0.143, p = 0.787; T3: 0.406, p = 0.425; mean: 0.543, p = 0.266). However, when considering the proportion of treatment sessions with a frustration level of 3, there was a strong and significant positive correlation with CAT scores at T3 (rho = 0.857, p = 0.029). Correlations with T2 and average scores were not significant (T2: rho = 0.068, p = 0.899; mean: rho = 0.541, p = 0.269).

For the KiddyCAT, small to moderate positive correlations were observed with mean frustration ratings, but none of these were significant (T2: rho = 0.319, p = 0.538; T3: rho = 0.359, p = 0.553; mean: 0.400, p = 0.505). Similar to the CAT analyses, when considering the proportion of sessions with a frustration level rating of 3, a strong positive correlation was obtained at T3, although this failed to reach significance (rho = 0.860, p = 0.061). Correlations with KiddyCAT scores at T2 and the mean across timepoints were moderate to strong but were also nonsignificant (T2: rho = 0.678, p = 0.139; mean: rho = 0.783, p = 0.118).

Research Question 5: Change following a Brief Intervention Period

Figure 5: KiddyCAT Scores across timepoints
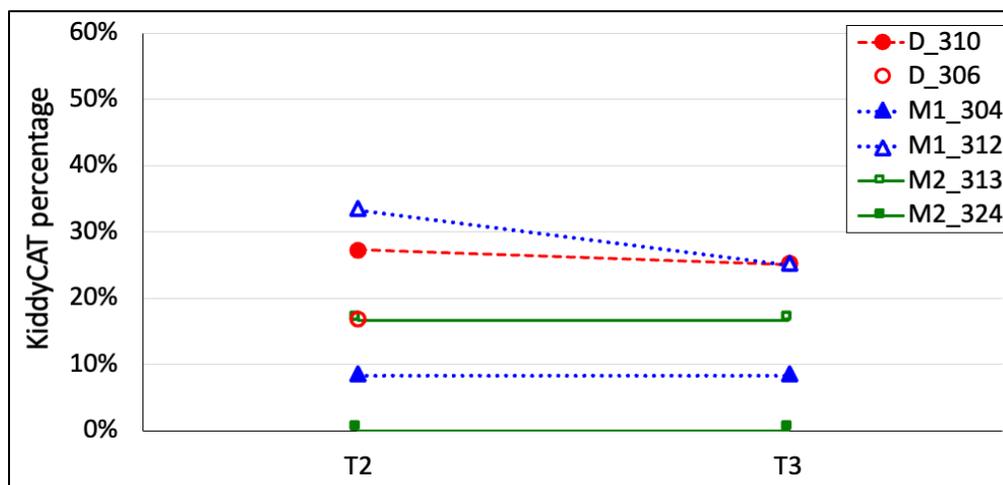
Figure 6: CAT Scores across timepoints
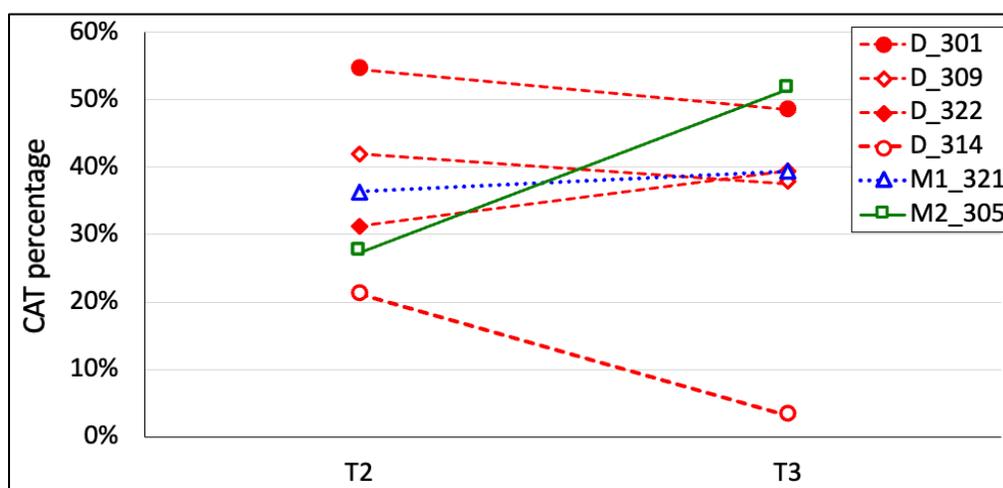
For younger children (KiddyCAT), the majority of scores remain the same across timepoints. One score in the massed-immediate group decreased from T2 to T3 (P312). One child who completed the KiddyCAT was assigned to the distributed group; that participant's score slightly decreased between timepoints (P310).

For the older children (CAT), all scores varied from T2 to T3. In the distributed group, scores of three participants decreased (P301, P309, P314) and one participant's score increased

(P322). Only one child who completed the CAT was assigned to the massed-immediate group; that participant's score increased slightly from T2 to T3 (P321). One child who completed the CAT was assigned to the massed-delayed group; that participant's score showed the greatest increase from T2 to T3 (P305, 27% to 52%).

DISCUSSION

This study was the first to examine self-reported attitudes from children with CAS about their speech, by administering the CAT and KiddyCAT to a group of children with CAS. By relating scores on these assessments to severity of diagnoses, parents' perceptions of participation across contexts, and average frustration ratings during therapy, a more comprehensive understanding of how-to best support children with CAS is gained. Because of the novelty of this research, scores that fell outside one standard deviation of the normative range were identified as possible negative attitudes.

Research Question 1: Communication Attitudes of Preschool Children with CAS

As a group, young children with CAS do not appear to feel negatively about their speech. This is on par with previous literature regarding the communication attitudes of young children who stutter (Guttormsen et al., 2015) and who have phonological disorder (McCormack, McLeod, & Crowe, 2019). Though preliminary, this finding is reassuring. Differences in type and consistency of impairment were expected to translate to negative attitudes; yet 5 out of 6 participants at T2 and 5 out of 5 participants at T3 indicated feeling more positive than negative about their communication. It is worth noting that one child's score at T2 (P312) fell above one

standard deviation of the normative range. At T3 the child's score was within norms. Alone, the T2 score is not indicative of a negative speech attitude but warrants further investigation.

There were several KiddyCAT questions where all answers indicated positive speech attitudes. At T2 and T3, all participants answered *yes* to the question (Q2), "Do you think you talk right?" It is possible that this relates to age; younger children may not fully understand their diagnosis or recognize the reason for going to therapy. Regardless, the consistency of this answer across timepoints is helpful for researchers, clinicians, and families to know. Participation in intensive speech therapy did not seem to affect young children's perception of the 'rightness' of their speech.

Questions commonly answered at individual timepoints were also identified. At T2, all children answered *no* to the question (Q4) "Do you think people need to help you talk?", which is a further indication that participation in speech therapy did not influence young children's concerns about their speech. At T3, all participants answered *yes* to Q3, "Do mom and dad like how you talk?", indicating that children were not equating parental concern regarding speech (i.e., putting the child in speech therapy) to their parents disliking how they talk. Additionally, all children answered *yes* to Q7, "Do you talk well with everybody?" This question relates to children's participation levels across contexts and indicates that young children may not feel their diagnosis impacts their ability to communicate with different people. The final two commonly answered questions both relate to if children think speaking is "difficult" (Q8) and whether words are "hard" to say (Q11). Of the KiddyCAT questions, Q8 and Q11 are the most demonstrative of awareness, or lack thereof, of speech difficulties. By the end of treatment all younger children did not think "talking is difficult" or that "words are hard for them to say."

Communication Attitudes of School-Age Children with CAS

Scores of school-age children with CAS are suggestive of more negative communication attitudes as compared to normative values and compared to the younger children. Half of the children who completed the CAT scored more than one standard deviation above the normative range at both timepoints. Two additional children fell above the normative range at T3. Prior research on children with cleft palates (Havstam et al., 2011), children who have articulation disorder (Luc & Brutten, 1990), and children who stutter (Vanryckeghem & Brutten, 2007) has assumed that older children become more aware of speech differences and individual abilities once they enter school, and therefore develop more negative speech attitudes. While this may be the case for children with CAS, it is also worth considering that differences in impairment and treatment may be influencing factors. CAS impairments are unpredictable, which can make children more difficult to understand, interfere with normal conversations, and make the disorder hard to treat.

There were several CAT questions where all answers indicated positive speech attitudes. At both timepoints, all children responded *false* to Q1 "I don't talk right," and *true* to Q7 "I like the way I talk." At T2, all children answered *false* to Q27 "I am not a good talker" and Q28 "I wish I could talk like other children." These answers indicate that when asked directly, school-age children with CAS feel like there is nothing wrong with how they speak. At T2, 5 of the 6 children answered *true* to Q9, "My parents like the way I talk" and at T3 all six children responded *true* to that question. This parallels with younger children's responses to the question, "Do your parents like how you talk?" and is an assuring sign that both older and younger children do not feel their parents think negatively of their speech because of their diagnosis.

Q18: "Other kids wish they could talk like me," was the only question in which all question answered *false,* indicating a negative speech attitude. This question will be further discussed in following sections regarding its applicability to overall attitudes.

Research Question 2: CAS Severity and Speech Attitudes

When CAT and KiddyCAT assessments were related to assessments of severity, findings were mixed. No significant correlation was found between the speech attitudes for school-age children with CAS and their DEMSS score. Increasing severity of diagnoses was expected to be associated with negative speech attitudes. For the KiddyCAT, a significant negative correlation was identified, meaning that preschool age children with CAS's attitudes about their speech are correlated with the severity of diagnoses. This finding, though preliminary, is critical. Clinicians and families informed of a possible correlation between severity of CAS and negative speech perceptions will be better able to provide necessary support for the child. For one child (P312), this correlation helps contextualize a borderline KiddyCAT score. This participant scored just above one standard deviation of the normative range at T2, and on the DEMSS, had significant evidence of CAS (a score of 323 or less).

Research Question 3: Caregiver Perceptions of Participation and Communication Attitudes

Though designed to assess Level (2) of the ICF: life participation, the FOCUS-34 is often used as a proxy for understanding the influence of communication disorders on Level (3) of the ICF: personal factors. Identifying if this holds true for children with CAS was of great interest. In this study, caregiver perceptions of participation across contexts were not significantly correlated with children's attitudes about their speech. Low statistical power due to a small sample size may

have occluded the detection of a significant correlation between these assessments. If this pattern

remains true with a larger sample size, it will indicate that these assessments measure two

distinct constructs - meaning parents' perceptions of participation across contexts are not

accurate indicators of how children with CAS feel about their speech. It is worth noting that a

study of young children with phonological disorder also found no significant correlation of

parental perception of participation across contexts and children's communication attitudes

(McCormack, McLeod, & Crowe, 2019). In conjunction, both findings suggest caregiver ratings

of child participation are not adequate substitutes for information gained from self-report

assessments of communication attitudes.

Research Question 4: Frustration During Therapy and Communication Attitudes

Higher average frustration ratings during therapy for both preschool and school-age

children with CAS did not correlate with negative speech perceptions. However, at T3, the

number of sessions in which school-age children were clearly frustrated and noncompliant (a

score of 3) did correlate with negative communication attitudes. Marked frustration during

sessions could have been due to several reasons (i.e., general unhappiness, tiredness, boredom,

stress, etc.). Higher levels of frustration during treatment could have exacerbated negative

attitudes about speech abilities. Due to the novelty and intensity of this treatment approach, this

is an important consideration. Given the small sample size and lack of findings at T2 or for

average scores, more research is needed before drawing further conclusions.

Research Question 5: Change in Scores over a Brief Intervention Period

When younger children's scores were compared across timepoints, no trends emerged. KiddyCAT scores remained the same at T2 and T3 for the majority of participants. Similarly, when treatment groups were compared, no trends emerged. Whether the child received treatment in the first two weeks, last two weeks, or throughout all four weeks of the study did not impact communication attitudes for younger children. This implies that the ASSIST protocol and camp format did not negatively or positively change how young children viewed their speech.

For older children with CAS, there was more variation in scores across timepoints and treatment groups. Scores of children in the massed-immediate and massed-delayed groups increased from T2 to T3. In particular, the score of the singular child in the massed-delayed group increased substantially between timepoints. This may mean that the intensive treatment that this child received between timepoints heightened awareness of any communication differences. Scores of those who received distributed treatment both increased and decreased, signifying that intensity of treatment was not a good predictor of changes in communication attitudes. Further research is needed before determining if this treatment protocol impacts how children view their speech.

Limitations

As the first investigation into communication attitudes as self-reported by children with CAS, there were a number of limitations to this study. First, this study had a small sample size; the CAT and KiddyCAT were only administered to a total of 12 children with CAS. This increased the risk of Type II errors due to low statistical power. Second, only children with CAS were included in this study, meaning that scores were only compared to established norms and

not current peers. Third, no communication attitudes assessments were administered at the first data collection timepoint (T1); scores may have already been influenced by the first two weeks of camp attendance. Finally, administrative errors required that some questions be excluded from analyses, which is a deviation from typical use of these assessments.

Future directions

Future research should include a larger sample size of children with and without CAS. Should these assessments be incorporated in future treatment studies for children with CAS, they should be administered at all data collection timepoints. On future distributions of the CAT and KiddyCAT, all questions should be included during analysis.

Wording on specific CAT questions should be reconsidered for future studies. During test administration, participants had difficulty understanding the meaning of Q2: "I don't mind asking a teacher the question in class." For young children, using ambiguous language like "I don't mind" is not ideal. For example, one participant answered *false, I don't mind,* and another participant responded *true, I don't mind,* but upon additional cueing, both meant that they were had no reservations about asking the teacher a question in class. Future revisions should consider changing ambiguous language like "I don't mind…" to wording more suitable for young children like "I'm okay with…." The connotation remains the same, which means that scoring does not change.

Question 18 on the CAT, "Other kids wish they could talk like me", has questionable translation to overall speech attitudes. In this study, all school-age children with CAS indicated that they did not think that other children wanted to talk like they do. Thinking that others do not want to talk like them, however, does not necessarily denote a negative speech attitude. If future

research distributes the CAT to children with and without CAS, it would be of interest to understand how typically developing children respond to this question and the underlying reasons for those responses.

It is also of interest to understand why this test requires true/false responses. Though school-age children can understand true and false statements, it would be clearer for the CAT to adopt yes/no statements, like the KiddyCAT. In this study, children often responded with yes/no or right/wrong despite being directed to respond true/false. This is a particularly important consideration when discussing future administration of the CAT to larger samples of children with CAS, as many of those children are at in increased risk for comorbid language disorders.

Future studies should also consider including a qualitative component. Several participants volunteered comments regarding their speech perceptions during data collection that provided context that quantitative assessments cannot provide. For instance, one participant (P314) made various remarks regarding his speaking improvements since the beginning of camp including "I'm working on [speaking] and getting better at it" and remarked that reading out loud in class "was hard but now it's easy." Incorporating a qualitative component in future studies of children with CAS will help provide reasons as to why children feel the way they do.

Given that several problems arose when the KiddyCAT and CAT were administered to children with CAS, future research would benefit from the development of a new self-report speech attitudes measure designed for children with SSDs. This measure should include questions better geared towards SSDs and be adaptable for administration to children with comorbid disorders.

CONCLUSION

Because of this study, researchers and clinicians can begin to understand how children with CAS feel about their speech. Understanding communication attitudes is critical - for many children, living with CAS means more than disrupted intelligibility or poor sound accuracy. Negative attitudes may impact interactions with peers, participation in school activities, or progress in treatment. When given the opportunity, children with CAS can provide credible, meaningful perspectives. In this study, scores of more than half of school-age children with CAS were indicative of negative speech attitudes. Though preliminary, these findings have clinical and familial implications. A speech-language pathologist who identifies a child with CAS to have negative speech attitudes may more adeptly create relevant treatment goals. Families who are aware of their child's attitudes will be better able to provide psychological support. Including communication attitude measures in future studies is a critical step towards understanding how to best treat and support children with CAS.

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