

**Critical Review:  
Literacy Outcomes in Children with Childhood Apraxia of Speech**

Jeness Weisgerber

M.Cl.Sc (SLP) Candidate

University of Western Ontario: School of Communication Sciences and Disorders

This critical review examined the current body of research investigating the literacy outcomes of children with childhood apraxia of speech. A search of electronic databases yielded eight studies with several designs, including case-control, longitudinal, single group, and case study. Overall, the evidence compiled here strongly indicates that children with childhood apraxia of speech exhibit reduced outcomes in areas of literacy development. Future research directions and clinical recommendations are provided.

***Introduction***

Childhood apraxia of speech (CAS) is a motor speech disorder that affects a child's ability to plan and program the movements needed to produce speech (American Speech-Language-Hearing Association (ASHA), 2007). Children with CAS often exhibit inconsistent speech errors that involve distortion of sounds as well as problems with syllable and/or word stress (ASHA, 2007). Therefore, these children may be highly unintelligible. CAS is a disorder that persists throughout childhood and even into adulthood, and generally requires intensive intervention in order to mitigate its effects (ASHA, 2007).

Due to the severe speech deficits exhibited by children with CAS, current treatments focus primarily on developing speech production through the principles of motor learning (Gillon & Moriarty, 2007). However, evidence shows that children with speech impairments are at an increased risk for developing literacy difficulties (Lefebvre et al., 2017). Gillon and Moriarty (2007) argue that children with CAS are a unique population due to the combination of motor planning and phonological awareness deficits faced by these children—deficits that have the potential to lead to more severe issues in literacy development. Moreover, research points to a high incidence of comorbid language disorders in children with CAS (Ekelman & Aram, 1983; Murray et al., 2018). The fact that children with CAS often struggle with both speech and language further highlights their increased risk for subsequent literacy difficulties (Gillon & Moriarty, 2007). While motor speech production issues are a critical intervention target for children with CAS, the potential for deficits in areas of literacy development could be detrimental to the future success of these children, and therefore, should not be disregarded.

***Objectives***

The primary objective of this review was to critically evaluate the current body of research regarding the literacy outcomes of children with CAS. The secondary objective was to inform clinical practice of professionals working with this population especially in terms of choosing targets for intervention.

***Methods***

**Search Strategy**

Articles included in this critical review were found on the computerized databases PubMed, CINAHL, and Education Database. The following key words were used: (childhood apraxia of speech) OR (developmental apraxia) OR (verbal dyspraxia) AND (literacy) OR (reading) OR (phonological awareness). Reference lists of searched articles were also examined to find additional applicable studies.

**Selection Criteria**

Studies chosen to be included in this critical review examined the outcomes of children with CAS in regards to both broad literacy outcomes as well as performance in specific areas of literacy development, such as phonological awareness. Studies that examined outcomes in areas known to be related to literacy development, such as morphological awareness, were also included.

**Data Collection**

The literature search identified eight studies pertaining to this topic, with the following research designs: four case-control studies, one longitudinal study, two single group, post-test only studies, and one descriptive case study.

## **Results**

### Case-Control Studies

Case-control studies are comparative group studies that examine correlations between a specific group and outcomes or traits. In these studies, the groups are predetermined based on the presence or absence of the disorder or condition in question, and therefore, do not include randomization. Though this research design has important limitations, such as the inability to determine cause and effect, it is useful when dealing with small sample sizes (Crandon, 2017). As such, it is a common design in studying communication disorders.

**Lefebvre et al. (2017)** conducted a case-control pilot study to examine the utility of a unique emergent literacy assessment protocol for children with CAS. This protocol focused on a broad range of emergent literacy skills, including phonological awareness, print awareness, and handwriting skills. This assessment included five standardized assessment measures and one parental questionnaire.

Participants were found using a convenience sample from a local preschool speech and language program and included eight children with suspected CAS (sCAS) and eight typically developing (TD) peers. All participants were four to five years of age and matched for age and sex. Inclusion criteria for study participation were clearly outlined. To be included in the sCAS group, participants had to be a) identified as having sCAS by their speech-language pathologist (SLP) b) identified as having normal receptive language skills according to standardized testing and c) experiencing speech sound production deficits. To be included in the TD group, participants had to have no academic, speech, or language concerns and no history of assessment or intervention in these areas, as determined by parent and teacher report. Each child participant underwent a 90-minute assessment of emergent literacy skills with a certified SLP. The parental questionnaire was administered to parents over the phone by a trained research assistant.

Appropriate statistical analysis of questionnaire results indicated that parents of TD children judged their children to be more advanced in phonological awareness and response to print in the environment than those parents of children with sCAS. Results from the standardized assessment measures found that children with sCAS scored significantly lower than their TD peers on tests of rhyme awareness, print awareness, letter knowledge, hand grasp, and motor coordination.

Strengths of this study included clear outlining of procedures and assessment protocols, resulting in high

replicability, as well as the use of standardized measures. Large effect sizes also added strength to this study. Weaknesses of this study were primarily related to the inclusion criteria used for the sCAS group. Diagnostic criteria for CAS, generally cited in previous research, were not used in this study. Furthermore, additional testing done on this sCAS group, for descriptive purposes, revealed that only two of the sCAS participants actually met the criteria for a diagnosis of CAS, based on guidelines laid out by McNeill et al. (2009b). Inclusion of a standardized measure in order to confirm absence of speech or language deficits in the TD group would have also increased the validity of this study. Another limitation of this study was the lack of data on previous experience with emergent literacy intervention for children in the sCAS group. Overall, this study provided somewhat compelling evidence that preschool age children with CAS demonstrate reduced outcomes in areas of emergent literacy development.

**Marion et al. (1993)** conducted a case-control study examining the rhyming outcomes of four children with CAS and four TD children, aged five to seven years. Four children were recruited to the TD group, from a larger participant pool, and were matched for age and sex. Inclusion criteria was clearly outlined for participation in either group.

Researchers administered four different rhyming tasks in order to measure the participants' skills in spontaneously generating rhyming words and judging the appropriateness of a rhyme in word pairs. The first task consisted of asking the child to produce a list of words that rhymed with a target word. The second task asked the child to determine the word that best rhymed with the target word, from a pair of two words. The third task asked the child to identify the words in a spoken list that rhymed with a target word. The fourth task involved making comparisons between acoustic features of vowels. Appropriate statistical analysis for the small sample size was used to determine that the children in the CAS group performed significantly more poorly than the children in the TD group across all rhyming tasks.

Strengths of this study included replicability and the use of a consistent method to classify children with CAS, as seen in previous literature. The addition of randomization in choosing from the participant pool of TD peers also adds to the validity of this study. Important limitations included the small sample size and the inability to account for confounding variables, due to the lack of matching between participant groups regarding language abilities. Overall, this study provided compelling evidence that children with CAS

exhibit deficits in early literacy-related rhyming outcomes compared to TD peers.

**McNeill et al. (2009b)** conducted a case-control study aimed at investigating differences in literacy outcomes between a group of 12 children with CAS and a group of 12 children with inconsistent speech disorder (ISD), matched for parameters of speech severity and inconsistency. ISD is a phonological speech disorder most often characterized by inconsistent speech productions without prosodic errors (McLeod & Baker, 2017). This study also included a third comparison group of 12 TD children. The three groups did not differ significantly in age (all between four and eight years old), gender, socio-economic status, or receptive vocabulary skills. Inclusion criteria and the participant selection process were very clearly outlined.

The authors utilized four standardized tests and two non-standardized informal tasks to examine outcomes in phonological awareness, letter-sound knowledge, underlying phonological representations, and real/non-word decoding. Due to the relatively large range of ages amongst participants in this study, some assessment measures were only used for particular age groups. For phonological awareness, two different standardized tests were used in order to ensure age-appropriateness. Furthermore, the two decoding tasks (real and non-word) were only administered to participants six years of age or older in each group. All assessments were conducted by trained SLPs, and mean inter-rater agreement for transcription of speech data was determined to be high (88.19%). There was also a high mean inter-rater reliability (93.4%) for the non-word decoding task.

Appropriate statistical analyses were employed to analyze results from the assessments. Large group effect sizes were found for phonological awareness, letter knowledge, and phonological representation outcomes. Regarding phonological awareness outcomes for the five-to-eight-year-old participants, the TD group performed significantly better than both the CAS and ISD groups, and the ISD group performed significantly better than the CAS group. For the four-year-old participants, the TD and ISD groups performed significantly better on phonological awareness compared to the CAS group. For letter knowledge and phonological representation, the TD group performed significantly better than the ISD and CAS groups. The reading (decoding) measures were qualitatively analyzed due to the small number of participants in the sample that were older than six years of age. Children in the CAS group had a lower overall range of percent consonants correct on the non-word reading task than the ISD and TD groups. Comparisons to norms on the

standardized measures also revealed that fewer participants from the CAS group fell within or above average range compared to the ISD or TD groups for phonological awareness, letter knowledge, and word reading measures.

An important strength of this study was the matching of the CAS and ISD groups for speech severity and inconsistency, as well as controlling for receptive vocabulary skills. Other strengths included the addition of a third comparison group, the use of standardized tests with strong psychometric properties, and good inter-rater reliability. One weakness of this study was the small sample size of children aged six years and older, which resulted in a lack of statistical analysis for reading outcomes. This weakens the generalizability of these results. Overall, this study provided compelling evidence that children with CAS demonstrate reduced outcomes in literacy development compared to their same age peers with ISD who exhibit similar speech severity.

**Miller et al. (2019)** conducted a case-control study comparing reading outcomes for a group of 40 school-age children with a history of sCAS to a group of 119 children with speech-sound disorder (SSD). The authors sought to a) determine the underlying speech and language deficits correlated to decoding issues in children with sCAS and b) determine predictors of reading proficiency in both groups. They divided both the sCAS and SSD groups into smaller subgroups of low-proficiency and average-proficiency readers.

Participants were chosen from a well-established longitudinal reading study, which consisted of children aged 7-18 years who had SSD or sCAS. Inclusion criteria for the original reading study were clearly outlined, as were the criteria for both groups in the current study. Diagnosis of sCAS was confirmed by two certified SLPs working on the original study. Individual testing occurred over two sessions of about three hours each. Various tasks from standardized tests were administered to determine outcomes in oral language, phonological awareness, decoding, rapid automatic naming, diadochokinetic (DDK) rates, single-word speech articulation, nonsense word repetition (NWR), multisyllabic word repetition (MSW), and performance IQ (PIQ).

Appropriate statistical analyses were utilized to determine that there was a significantly higher percentage of low-proficiency readers in the sCAS group compared to the SSD group. The low-proficiency CAS group exhibited reduced outcomes compared to the average-proficiency CAS group on measures of phonological awareness, language, NWR, and DDK

rates. In comparing the sCAS and SSD groups, the average-proficiency readers from the sCAS group were found to score significantly lower than the average-proficiency readers from the SSD group on measures of NWR and DDK rates. Additionally, the low-proficiency readers from the sCAS group scored significantly lower than the low-proficiency readers from the SSD group on measures of NWR, DDK rates, and single-word articulation. Using multivariate logistic regression analysis, significant predictors of low reading proficiency in both disorder groups were determined to be oral language, phonological awareness, MSW, and DDK rates, but not group membership.

Strengths of this study included the large sample size, not often seen in CAS studies, as well as the evidence-based criteria used to determine inclusion in the sCAS group. Limitations included the lack of information about matching of participants and the failure to control for extraneous variables, such as history of speech, language, or literacy intervention. Overall, this study provided compelling evidence that children with CAS exhibit reduced outcomes similar to peers with SSD in areas related to literacy development, such as phonological awareness, decoding, and language. It also provided strong evidence that children with CAS exhibit reduced outcomes in other areas, such as NWR and motors-speech measures, compared to peers with SSD.

#### Longitudinal Studies

Studies with a longitudinal design collect data at multiple points over an extended period of time and as such, these types of studies are valuable for examining how disorders progress over time (Institute for Work & Health, 2015). Therefore, they are especially informative when researching outcomes of developmental communication disorders in children.

**Lewis et al. (2004)** conducted a longitudinal study that compared speech and language outcomes for three groups of school-age children: 10 with CAS, 15 with other SSDs, and 14 with combined speech-sound and language (SL) disorders. The children were followed from preschool age until 8-10 years old. The authors aimed to compare outcomes on specific measures of articulation, DDK rates, language, reading, and spelling. Assessments administered at preschool age included three standardized tests and three non-standardized measures. Assessments administered at school-age included six standardized tests and four non-standardized measures.

Appropriate statistical analysis found significant main effects for group on all preschool measures, with moderate to large effect sizes. The CAS group had poorer outcomes than the SSD group on all measures;

however, these measures did not differ between the CAS and SL disorder groups at preschool age. Except for one articulation measure, significant main effects for group were also found on all school-age measures, with small to large effect sizes. At school-age, the CAS group was revealed to have poorer outcomes than the SSD group on all measures, and also had poorer outcomes than the SL disorder group on NWR, DDK rates, PIQ, language, and spelling. Further statistical analysis (ANCOVAs, post-hoc tests) was used to determine the residual change (change from preschool to school-age) for each group. Significant main effects for group were found on measures of language and NWR, with small to moderate effect sizes. Overall, the CAS group demonstrated less progress from preschool to school-age in regards to language measures than the other two groups.

One notable strength of this study included the longitudinal design, which provided valuable information about the outcomes of CAS over time, including degree of change. Other strengths included the clearly described and evidence-based inclusion criteria, as well as the use of several standardized assessment measures. The inclusion of a third SL disorder comparison group was also an important strength of this study. Weaknesses of this study included the failure to control for extraneous variables, such as speech severity, receptive language skills, and history of speech therapy. Furthermore, even though several standardized tests were used at both age points, the tests administered at preschool versus school-age differed greatly. Maintaining these assessment measures across ages would have increased the validity of this study's findings. Overall, this study provided compelling evidence that children with CAS demonstrate poorer outcomes in specific areas of language and literacy development than their same-age peers with SSD and SL disorder, and that these deficits may persist into school-age.

#### Single Group, Post-Test Only Studies

Single group, post-test only studies involve analyzing outcomes from one group. As such, this type of study does not include any control or comparison groups, which makes it a weaker study design (Price et al., 2017). However, when no comparison group is available, this type of study can provide preliminary research results.

**Marquardt et al. (2002)** completed a single group, post-test only study examining the syllable manipulation skills of children with CAS. Participants were three children with moderate to severe CAS. A group of three TD children was also included in this study strictly as a measure by which to validate the age appropriateness of

assessment tasks. Inclusion criteria for all participants were clearly outlined.

During this study, three tasks were administered in order to examine the participants' ability to detect syllables and judge intrasyllabic position and structure. In the syllable detection task, the participants were asked to tap out the number of syllables in the word. In the judgement tasks, participants were asked to make judgements about phonemes within syllables using colored blocks as cues. All tasks began with demonstrations and screening items, and the participants were administered the experimental items only if they were first able to answer a certain number of the screening items correctly. All experimental items were provided in random order. The researchers did not employ any statistical measures for data analysis, but instead relied on the raw number of correct responses for a descriptive analysis. Results revealed that all three of the participants in the CAS group demonstrated difficulty across syllable tasks, despite having participated in speech therapy for at least three years.

One strength of this study was the replicability of study procedures. This included a clear explanation of the three syllable tasks, which was particularly important for these non-standardized procedures. The randomization of test item order also increased the validity of results. This study had some important limitations, such as the small sample size, the lack of a comparison group, the use of non-standardized assessment tasks, and descriptive analysis rather than statistical analysis. Due to these limitations, it is difficult to generalize these results, particularly to children with mild CAS. Therefore, this study provided suggestive evidence that children with moderate to severe CAS exhibit reduced outcomes on syllable tasks.

**Murray et al. (2018)** completed a single group, post-test only study examining the morphological skills of 26 children with CAS, four to five years of age. They sought to determine the extent to which morphological deficits in children with CAS could be explained by motor speech deficits. Participants were chosen from a pool of 59 children taken from another CAS study. Inclusion criteria were clearly outlined (e.g., aged 4;0-5;11 years, met criteria for diagnosis of CAS, no diagnosed comorbid developmental or genetic disorders) to determine the 26 participants who met the current study's requirements.

Participants in this study were assessed using the Clinical Evaluation of Language Fundamentals – Preschool – Second Edition (CELF-P2; Wiig et al., 2006). In particular, the Receptive Language Index (RLI) and the Expressive Language Index (ELI), as well

as individual expressive language subtest scores, were analyzed. Scores revealed that 19% of participants fell within the range of a receptive language disorder and 50% of participants fell within the range of an expressive language disorder. An appropriate statistical analysis (ANOVA) was used to determine that ELI scores were significantly lower than RLI scores amongst the participants. On specific expressive language subtests, participants were found to perform significantly more poorly on subtests that involved manipulation of morphology or morphosyntax than on subtests that assessed expressive vocabulary skills. Further analysis of specific morpheme production revealed that most morphemes, except for “-ing”, failed to meet age expectations. Examination of speech productions for the presence of common CAS motor speech errors also revealed that many, but not all, morphological errors could be explained by motor speech deficits. Items for three random participants were coded by two authors, with a very high inter-rater reliability (94.8%).

The use of a standardized, norm-referenced test with good psychometric properties (CELF-P2) was an important strength of this study. This test is widely used in clinical practice, and therefore, its use also increased the clinical applicability of the results, particularly in regards to understanding the influence of motor speech deficits on test stimuli. High inter-rater reliability was also a strength. Limitations of this study were that data from two participants was unavailable for item analysis and the statistical power was limited with the small sample size. Overall, this study provided compelling evidence that some children with CAS exhibit morphological deficits that indicate a possible comorbid language disorder.

#### Case Studies

Case studies are nonexperimental research designs. They provide a detailed description of a patient, disorder, or treatment. Though they do not provide a strong level of research evidence, they are sometimes the only option in instances of less common communication disorders, and can contribute to further areas of research (Price et al., 2017).

**Zaretsky et al. (2010)** conducted a case study of an 11-year-old girl (“LH”) with severe CAS and borderline IQ (Full Scale Intelligence Quotient = 74). The researchers outlined the speech, language, cognitive, and literacy outcomes of the participant, as well as her therapy progress. LH's background was clearly described, including her performance on various measures and therapy history from 6-11 years of age. LH received several years of intervention focusing specifically on phonological awareness, phoneme-grapheme mapping,

and reading comprehension. Despite gains in her reading comprehension (falling within the average range), LH continued to struggle with phonological awareness and decoding. The authors assumed that her speech deficits were greatly contributing to these struggles. As such, a specialized treatment program, focusing on both speech and literacy, was administered in order to address these continued deficits. The researchers sought to further examine her phonological awareness and language outcomes by administering several assessment measures at the end of therapy. They administered five standardized tests and two non-standardized tasks in order to determine outcomes in language, automaticity and accuracy of retrieval, syllable and phoneme manipulation, phonological and working memory, decoding and encoding, and reading (rate, fluency, accuracy, comprehension). For the two non-standardized tasks, responses were transcribed and scored by a second observer, with high inter-rater reliability.

At the end of therapy, LH demonstrated improvements in syllable segmentation and vowel identification. She also made progress in decoding skills, though this was less stable, and non-word decoding continued to be a greater challenge. Outcomes of the standardized assessment measures indicated ongoing language disorder. Assessment also revealed ongoing deficits in phonological awareness and decoding and encoding, while reading fluency, accuracy, rate, and comprehension were all within average range. Non-standardized tasks also revealed deficits in NWR (phonological memory), and word recall (working memory), with working memory outcomes being significantly worse than a group of similar children with language impairment (no CAS).

The detailed description of longitudinal participant data and the clearly outlined study procedures, allowing for easy replication, were strengths of this study. The authors also included clear justification for all measures used. This, along with the use of several standardized tests, offers validity to this study. Limitations included the inherent nature of a case study design, which is descriptive and does not allow for generalizability. In particular, the participant in this study had severe CAS and borderline IQ, making the results difficult to extrapolate to cases of mild or moderate CAS with average IQ. Overall, this study provided somewhat suggestive evidence that children with severe CAS and borderline IQ exhibit reduced literacy and literacy-related outcomes.

## ***Discussion***

This critical review examined research in the area of literacy outcomes for children with CAS. Though the level of evidence provided by these studies was somewhat mixed, taken together, they provided compelling evidence that children with CAS demonstrate reduced outcomes in literacy development. Amongst these studies, several trends were found in specific areas of literacy development.

Six out of the eight studies examined in this review provided suggestive to compelling evidence that children with CAS exhibit reduced outcomes in areas of phonological awareness (Lefebvre et al., 2017; Marion et al., 1993; Marquardt et al., 2002; McNeill et al., 2009b; Miller et al., 2019; Zaretsky et al., 2010). These findings are significant, as we know that phonological awareness skills are crucial to the foundation of literacy skills (Gillon & Moriarty, 2007). Two of these studies (Lefebvre et al., 2017; McNeill et al., 2009b) also provided strong evidence that children with CAS exhibit deficits in related areas of emergent literacy development, including print awareness and letter knowledge. Most notably, McNeill et al. (2009b) provided compelling evidence that children with CAS exhibit reduced outcomes in phonological awareness compared to peers who have ISD and similar levels of speech severity. This suggests a particular phonological awareness deficit for children with CAS that is potentially more severe than children with other SSDs. Miller et al. (2019) failed to find a significant difference in phonological awareness outcomes between low proficiency readers with CAS and those with other SSDs. However, the evidence provided by this study is slightly less compelling, as the researchers did not control for speech severity.

Four studies found that children with CAS exhibit reduced outcomes in decoding (Lewis et al., 2004; McNeill et al., 2009b; Miller et al., 2019; Zaretsky et al., 2010). Two of these studies also found evidence of spelling deficits (Lewis et al., 2004; Zaretsky et al., 2010). Overall, these studies provided compelling evidence for deficits in decoding and encoding amongst children with CAS. Most compellingly, McNeill et al. (2009b) and Lewis et al. (2004) found reduced outcomes in word reading in comparison to same age peers with other SSDs, while Lewis et al. (2004) also found reduced outcomes in spelling compared to peers with SL disorder at school-age.

Two studies (Lewis et al., 2004; Miller et al., 2019) provided compelling evidence that children with CAS perform more poorly on NWR tasks than peers with other SSDs. Lewis et al. (2004) also found this outcome

compared to peers with SL disorder at school-age. In addition, Zaretsky et al. (2010) noted that LH demonstrated reduced outcomes in NWR. These findings are important because NWR tasks measure phonological short-term memory and deficits in this domain have been linked to reduced reading abilities (Baird et al., 2011). For children with CAS, it can be difficult to determine if deficits in NWR are due to motor speech errors or issues with phonological processing, but given the complex speech and language profile of many children with CAS, we can assume that these deficits are due to a combination of these two factors (Miller et al., 2019).

Several of the studies included in this critical review examined language outcomes in children with CAS. Taken together, Lewis et al. (2004), Miller et al. (2019), Murray et al. (2018), and Zaretsky et al. (2010) provided compelling evidence that children with CAS exhibit reduced outcomes in regards to expressive and receptive language skills. Notably, Lewis et al. (2004) found that these deficits in receptive and expressive language persisted despite improved outcomes in articulation, indicating that improved speech may not equal a resolution of language deficits for children with CAS. Similarly, the participant in the Zaretsky et al. (2010) study continued to demonstrate deficits in language despite improvement in other domains. Though these findings are significant, both these studies examined outcomes for children with moderate to severe CAS, so generalizing these conditions to children with mild CAS should be done with caution. Murray et al. (2018)'s findings on the morphological skills of children with CAS are of particular interest for this review in that morphological awareness skills are an important foundation for the development of literacy skills (Wolter et al., 2009). Furthermore, the presence of morphological errors that were unexplained by motor speech errors serves to further strengthen the assumption that language deficits may persist despite gains in speech. Overall, the receptive and expressive language deficits exhibited in these studies are strongly indicative of the reduced literacy-related language outcomes of children with CAS.

Further research is needed to determine the specific areas of literacy development most impacted in children with mild to severe CAS, compared to peers with other speech-sound and language disorders. Future studies should control for extraneous variables such as speech severity and history of intervention in order to increase the validity of findings. Research regarding the efficacy of intervention protocols that integrate motor speech and literacy targets should also continue (see McNeill et al., 2009a; Moriarty & Gillon, 2006).

### ***Clinical Implications***

Overall, the studies reviewed here indicate that children with CAS experience deficits in areas of literacy development, such as phonological awareness, phonological memory, decoding and encoding, morphological awareness, and language. As such, clinicians who work with these children should be aware of these potential deficits in order to provide intervention as early as possible. Clinicians should also understand that literacy-related goals are important and appropriate intervention targets for children with CAS, and be careful not to assume that an improvement in speech production alone will lead to improvements in literacy outcomes. In order to address the exceptional needs of these clients within a limited therapy timeframe, it is possible that the best intervention protocol would focus on integrating both motor speech and literacy-related targets.

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