Research Article

Reading Outcomes for Individuals With Histories of Suspected Childhood Apraxia of Speech

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Purpose: The primary aims of this study were to examine the speech-language correlates of decoding difficulties in children with histories of suspected childhood apraxia of speech (sCAS) and to identify predictors of low-proficiency reading levels.

Method: Participants were school-age children and adolescents, 7–18 years of age, diagnosed with sCAS ($n = 40$) or speech sound disorder but no sCAS (SSD-no sCAS; $n = 119$). The sCAS and SSD-no sCAS reading groups were compared on measures of performance IQ, oral language, phonological awareness, rapid automatic naming, diadochokinetic rates, single word articulation, and multisyllable and nonsense word repetition. Logistic regression analyses were employed to identify predictors of low-proficiency reading in the sCAS and SSD-no sCAS groups.

Results: Sixty-five percent of the participants with sCAS compared to 24% of those with SSD-no sCAS were classified as low-proficiency readers based on nonsense and single word decoding. Analysis failed to reveal significant differences in reading, oral language, or phonological awareness between low-proficiency readers with sCAS and low-proficiency readers with SSD-no sCAS. Oral language and phonological awareness skills were the best predictors of reading level for all participants, followed by performance on multisyllabic word repetition and diadochokinetic rate.

Conclusions: The language and phonological awareness deficits of children with sCAS are related to their risks for reading failure. To a lesser degree, motor speech deficits and speech sound production also increase risks for reading difficulties. The findings justify early intervention for this subset of children.

Childhood apraxia of speech (CAS) is a neurologically based, pediatric speech sound disorder (SSD) whose underlying cause is thought to be related to difficulty in selecting and executing the motor plans for speech production (American Speech-Language-Hearing Association [ASHA], 2007). This difficulty with sequencing and producing speech sounds will often render a child's speech highly unintelligible. While the long-term consequences of CAS on speech intelligibility and articulation skill have been well studied (Aram & Nation, 1982; Crary, 1993; Hall,

Jordan, & Robin, 1993, 2006; Rosenbek & Wertz, 1972; Stackhouse & Wells, 1997; Velleman, 2016; Velleman & Strand, 1994), there is less research on school-age literacy skills in children with this disorder. Research on other forms of SSD, those with cognitive-linguistic bases, for example, suggests that poorly specified and organized phonological information, that is, a core phonological processing deficit, is common to both SSD and decoding difficulties (Anthony et al., 2011; Cabbage, Farquharson, Iuzzini-Seigel, Zuk, & Hogan, 2018; Pennington & Bishop, 2009; Rvachew & Brosseau-Lapre, 2018). There is less research, however, about how these same deficits impact decoding for children with severe and persistent speech disorders as in CAS.

The low prevalence of CAS may help to account for a paucity of research on decoding skills in this disorder. Its prevalence in the general population ranges from 0.1% to 0.2% or one to two children per thousand (Shriberg, Aram, & Kwiatkowski, 1997a) and is reportedly found in just 3.4%–4.3% of children referred for SSDs (Delaney & Kent, 2004). Research in communication disorders on relatively low-incidence conditions, such as CAS, may not have access

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to large enough participant samples to conduct randomized controlled trials, the gold standard of research (Justice, Nye, Schwarz, McGinty, & Rivera, 2008).

Another explanation for the lack of research about the association of CAS and decoding difficulties is that CAS has been viewed historically as primarily a disorder of motor speech control (Duffey, 2003; Robin, 1992; Shriberg et al., 1997a). A review of the literature, which includes the official definition of CAS by ASHA, evidences this circumpect view of CAS. This conceptualization of CAS as affecting speech sound production primarily belies the fact that children with CAS often present with a broad and significant range of language and literacy problems (Aram & Nation, 1982; Gillon & Moriarty, 2007; Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004; Marion, Sussman, & Marquardt, 1993; Marquardt, Sussman, Snow, & Jacks, 2002; McNeill, Gillon, & Dodd, 2009; Zaretsky, Velleman, & Curro, 2010). Finally, the defining characteristics of CAS, including the contribution of deficits in language and literacy, continue to be disputed as evidenced by its evolving nomenclature (ASHA, 2007; Aram & Nation, 1982; Cray, 1993; Hall et al., 1993, 2006; McCabe, Rosenthal, & McCloud, 1998; Morley, 1957; Orton, 1964; Rosenbek & Wertz, 1972; Shriberg et al., 1997a; Shriberg, Aram, & Kwiatkowski, 1997b; Velleman & Strand, 1994; Yoss & Darley, 1974).

CAS has been conceptualized in at least three different ways, each of which predicts difficulties with decoding. While it is beyond the scope of this article to evaluate the validity of differing views of CAS, these perspectives provide evidence for the complexity of the relationship between CAS and literacy skills. One conceptualization considers CAS a disorder of motor speech planning with language and related literacy difficulties viewed as co-occurring conditions rather than defining features (ASHA, 2007; Hall et al., 1993, 2006). ASHA's conceptualization of CAS is consistent with this first view. CAS is defined as a speech disorder in which the core impairment is in "planning and/or programming spatiotemporal parameters of movement sequences, resulting in errors in speech sound production and prosody" (ASHA, 2007, pp. 3–4). ASHA acknowledges the presence of co-occurring symptoms, including symptoms that represent the consequences of CAS and those that are considered "nonrelated" but co-occurring, such as language and learning disabilities, including reading disabilities (ASHA, 2007, n.d.). In this conceptualization, language and literacy difficulties might occur in the same manner and frequency as reading disorders in the general population (Hall et al., 1993, 2006). Conversely, these difficulties may be secondary consequences of the severe and persisting nature of the speech difficulties associated with CAS (Gillon & Moriarty, 2007; Stackhouse & Wells, 1997; Stoel-Gammon, 2011; Velleman, 2011). Decreased vocalization during infancy and subsequent consistently distorted speech have an "upstream impact on the cognitive–linguistic aspects of phonology," such as the establishment of accurate phonological representations that affect language and literacy development (Velleman, 2011, p. 83).

According to a second view of CAS, language difficulties are a more fundamental feature of the disorder. Deficits in linguistic skills and motor speech impairment are proposed to originate from a common underlying deficit in phonological representations that affect the simultaneous organization of language and speech (Aram & Nation, 1982; Marquardt et al., 2002; Nijland, 2009; Velleman & Strand, 1994). The "critical age hypothesis" proposes that the persistence of a speech disorder (as in CAS) at the time children are first learning to read undermines literacy development (Bird, Bishop, & Freeman, 1995; Bishop & Adams, 1990; Hayiou-Thomas, Carroll, Leavett, Hulme, & Snowling, 2017). A modified version of the "critical age hypothesis" links phonological processing skills to speech production and suggests that poorly specified phonological representations simultaneously cause persistent speech errors and undermine the development of phonemic awareness crucial for the development of written language skills (Nathan, Stackhouse, Goulondris, & Snowling, 2004). Children with severe and persistent speech problems, as in CAS, are thus more likely to struggle with literacy acquisition. Correspondingly, a diagnosis of CAS confers an increased risk of reading disorder.

A third conceptualization of CAS is as a motor speech disorder with a complex set of associated deficits arising from a heterogeneous genetic source (Chilosi et al., 2015; Worthey et al., 2013). In this conceptualization, CAS is not a unitary disorder with all children demonstrating the same set of symptoms (Nijland, Terband, & Maassen, 2015; Velleman, 2016); instead, children can evidence differing levels of severity of motor speech deficit and differing patterns of cognitive, language, and literacy deficits (Nijland et al., 2015; Peter, Button, Stoel-Gammon, Chapman, & Raskind, 2013; Peter et al., 2016; Worthey et al., 2013). Moreover, these features may change during the child's course of development (Lewis et al., 2004; Velleman, 2016). According to this view, language and reading difficulties can be considered part of the disorder's "symptom complex" (Nijland et al., 2015; Velleman, 2016). Independent genetic and environmental factors, both risk and protective, will determine the specific underlying deficit, and phenotypic differences in the disorder will depend on the combination of these liabilities that exceed a given threshold (Pennington, 2006; Pennington & Bishop, 2009). Genetic studies support a heterogeneous genetic basis for CAS with differing pathways and interactions, creating various phenotypes including reading and language impairment (Peter et al., 2013; Worthey et al., 2013).

These three conceptualizations acknowledge that children with CAS may be at increased risk for reading impairment relative to other forms of SSD. Consistent with the first conceptualization, poorer linguistic abilities (both global oral language and phonological processing abilities) and the ensuing reading and spelling deficits for school-age children with CAS may result from the cascading effect of motor speech difficulties on a child's developing language and literacy systems (ASHA, 2007; Stoel-Gammon, 2011; Velleman, 2011). Stackhouse and

Wells (1997) offer their account of the effects of motor speech impairment on language and literacy whereby poor speech output interferes with the accurate encoding of complex words, which affects the child's developing lexicon and linguistic system. There is evidence that, even for typically developing infants, less expansive speech production is associated with decreased letter identification at school age, a skill that has been consistently associated with decoding ability (Farquharson et al., 2018).

While some studies of children with SSD have failed to confirm the effects of poor speech production on phonological processing and literacy skills, these studies have not explicitly examined children with CAS. For example, Rvachew and colleagues (Rvachew, 2007; Rvachew & Grawburg, 2006) found that reading difficulties in children with SSD were more closely related to weaknesses in phonological processing than to speech impairment and that articulation did not have a direct effect on phonological awareness. Additional research by Peterson, Pennington, Shriberg, and Boada (2009) suggests that comorbid language impairment is more closely associated with reading ability than speech production difficulties. Findings from this study are consistent with previous research indicating that language impairment accounts for a significant proportion of the shared variance of SSD and reading disorder (Lewis, Freebairn, & Taylor, 2000). While it is likely that language impairment impacts literacy skills in a similar manner for both children with CAS and those with other forms of SSD, a study by Lewis et al. (2004) comparing school-age outcomes for children with CAS, SSD only (SSD), and SSD with accompanying language impairment (SSD + LI) demonstrated that the speech articulation and single-word decoding skills of the children with CAS were significantly reduced compared to the children with SSD and children with SSD + LI. The severity, persistence, and inconsistency of speech production difficulties observed in CAS may account for the greater trouble in decoding skills for these children.

In addition to the motor speech difficulties of children with CAS, the increased rates of impairments in language and language-related literacy skills observed in children with CAS relative to the population estimates may contribute to reading problems (Ekelman & Aram, 1983; Lewis et al., 2004; Thoonen, Maassen, Gabreëls, Schreuder, & de Swart, 1997; Tomblin et al., 1997). If reading difficulties were strictly related to motor speech problems and not additional language impairments, we would anticipate difficulties with expressive compared to receptive language skills. Expressive grammar deficits related to restrictions in motor speech skills or phonological simplifications would be expected to subside with improvement in speech production. That is, deficits observed in morphology due to the child's inability to produce (articulate) grammatical markers (e.g., plural endings *-s* or *-es*) would improve as soon as the child can articulate them. The results of the Lewis et al. (2004) study above that followed groups of school-age children recruited from a clinical population with CAS, SSD, or SSD + LI run counter to both expectations. The

study revealed high rates of impairment in both receptive and expressive language skills in the group with CAS with accompanying deficits in reading and spelling skills (Lewis et al., 2004).

Findings from Murray, Thomas, and McKechnie (2018) are consistent with previous research in demonstrating that some children with CAS have restrictions in expressive grammar that cannot be entirely explained by articulation or motor speech issues. For example, Ekelman and Aram (1983) found that participants with CAS evidenced expressive grammar errors unrelated to motor speech skills or phonological simplifications (i.e., errors in pronoun use, misuse of irregular past tense morphemes, omission of copulas in yes/no questions, and difficulties with question transformations). Thus, it appears that the persistence of motor speech difficulties, in conjunction with language impairments, contributes to decoding problems in children with CAS. What is not known is why some children with CAS present with average decoding skills while others demonstrate below-average decoding abilities.

Given the current conceptualization of CAS as a motor speech disorder with merely co-occurring or even secondary language and literacy deficits, the customary interventions rely primarily on the habituation of accurate motor speech plans (Gillon & Moriarty, 2007). However, if reading difficulties are viewed as a commonly occurring feature of CAS, similar to other SSDs, interventions designed for children with language and literacy problems are suitable for children with CAS. Moreover, literacy interventions that target motor speech difficulties, in conjunction with the morphological weaknesses that are specific to children with CAS, might also be encouraged (Gillon & Moriarty, 2007; McNeill et al., 2009; Murray, McCabe, & Ballard, 2014). Since many children with CAS have reading disabilities (Gillon & Moriarty, 2007; Lewis et al., 2004; Zaretsky et al., 2010), viewing CAS as primarily a motor speech deficit may delay the provision of preliteracy and literacy interventions until the child's speech has improved and thus stall earlier and potentially more effective reading interventions.

The primary goals of the current study were to assess the frequency of reading difficulties in a cohort of children with histories of suspected CAS (sCAS) and compare the cognitive-linguistic correlates of these difficulties to those of children who have SSD without sCAS (SSD-no sCAS). As there is a dearth of information about literacy skills in children diagnosed with CAS, we wished to examine the similarities and differences with another SSD known to have an association with reading ability. We grouped the participants into average-proficiency and low-proficiency reading groups. In addition, we conducted a regression analysis on continuous data to determine predictors of reading proficiency. We sought to answer the following three research questions:

1. How do children with sCAS with average-proficiency decoding differ from those with low-proficiency decoding on measures of speech- and language-related skills, including oral language, phonological awareness, rapid

automatic naming, diadochokinetic rates, single-word speech articulation, and nonsense and multisyllabic word repetition?

2. How do children with sCAS with average- and low-proficiency decoding differ from children with other forms of SSD (SSD-no sCAS) with average- and low-proficiency decoding on these same measures?
3. Which measures best predict decoding skills in children with sCAS and SSD-no sCAS?

We hypothesized that reading difficulties would be common in children with sCAS and that children with sCAS and average-proficiency decoding skills would differ from children with sCAS and low-proficiency decoding skills in speech severity and persistence, global oral language abilities, and phonological processing skills. An additional expectation was that global oral language abilities and phonological processing skills would be related to literacy skills in children with sCAS and those with other forms of SSD. Finally, we anticipated that continued speech production difficulties and poor motor speech skills into school age would confer additional risk for poor decoding skills for children with sCAS and those with other less severe forms of SSD.

Method

Participants

The current study utilized participants who were enrolled in the Cleveland Family Speech and Reading Study between 1990 and 2014. The Cleveland Family Speech and Reading Study is a longitudinal study that examined the genetic bases of SSD (Lewis et al., 2004). This unique cohort was assessed at multiple time points for speech, language, and literacy skills. Children were referred to the original study with a diagnosis of an SSD from the clinical case-loads of speech-language pathologists and had a range of severity of SSD, including sCAS.¹ For this study, participants were assessed at school age (7–18 years). A wide age range was established for the current study to include as many participants with sCAS as possible, as CAS is, as noted previously, a low-incidence disorder (Delaney & Kent, 2004; Shriberg et al., 1997a).

Inclusion Criteria for the Cleveland Family Speech and Reading Study

Study participants in the original study met the following general inclusion criteria based on information provided by a parent in an interview or via a questionnaire: normal hearing acuity; fewer than six episodes of otitis media prior to age of 3 years; monolingual English speaker; and absence of a history of neurological disorders other than CAS,

such as cerebral palsy or autism spectrum disorder. An additional criterion that determined eligibility into the study was documentation of normal intelligence as defined by a prorated performance IQ (PIQ) of at least 80 on the Wechsler Preschool and Primary Scale of Intelligence–Revised (Wechsler, 1989), Wechsler Intelligence Scale for Children–Third Edition (Wechsler, 1991), or Wechsler Intelligence Scale for Children–Fourth Edition (Wechsler, 2003). Eligible children were also required to pass a pure-tone audiometric screening at 20 dB HL bilaterally. The Four-Factor Index of Social Status survey was administered to assess family socioeconomic status (Hollingshead, 1975).

Inclusion Criteria for sCAS

A subset of children who were enrolled in the Cleveland Family Speech and Reading Study had a history of sCAS diagnosis (and treatment) from experienced speech-language pathologists. Upon entry into the study, this diagnosis was confirmed by an ASHA-certified speech-language pathologist serving on the research team with experience in motor speech disorders and reconfirmed by a second ASHA-certified speech-language pathologist serving on the study. Forty-two participants with sCAS qualified for the current study because they received an initial or follow-up assessment that included a test of reading skill, an assessment of phonological processing, and an assessment of global language ability that occurred between 7 and 18 years of age. Two children were randomly eliminated from the study, as they had a sibling who was also enrolled in the study. This was done to ensure independent observations, which resulted in a sample of 40 children with sCAS. The majority of the children who qualified for the current study were enrolled at preschool age (4–6 years; 31/40); a smaller number were enrolled at school age (7–11 years; 7/40) and adolescence (13–14 years of age; 2/40). For three of the older participants, this was an initial evaluation.

At the time of initial enrollment in the study, the diagnosis of sCAS by the referring speech-language pathologist was confirmed by requiring that the participants exhibit a minimum of four of the following seven features of CAS, as reported in the literature. These were observed in a conversational speech sample or by direct assessment:

1. greater difficulty with polysyllabic words than with monosyllabic words (syllabic integrity is affected by omissions of syllables and consonants and cluster simplification; Jacks, Marquardt, & Davis, 2006; Lewis et al., 2004; Marquardt et al., 2002; Rosenbek & Wertz, 1972; Velleman, 2003; Yoss & Darley, 1974);
2. restricted phonemic repertoire and reduction in syllable shape (Davis & Velleman, 2000; Overby & Caspari, 2015);
3. atypical phonological errors (e.g., initial consonant deletion, voicing and unvoicing errors, and glottal replacement; Lewis et al., 2004; Skinner-Meredith & Smith, 2013; Yoss & Darley, 1974);

¹The majority of this cohort was diagnosed with CAS in 1990–2003. The diagnostic criteria for CAS continue to evolve; therefore, to be conservative in our diagnoses, we are using the term *sCAS*.

4. a combination of two and three sound feature errors, prolongations and repetitions of sounds or syllables, and distortions, omissions, and additions in speech repetition tasks or conversational speech (Yoss & Darley, 1974);
5. abnormal prosody, slow rate, and equal stress especially in older children or those who have received speech therapy (Shriberg, Aram, & Kwiatkowski, 1997c; Yoss & Darley, 1974);
6. metathetic errors (Austin & Shriberg, 1996; Shriberg et al., 2003); and
7. reduced diadochokinetic rates often produced with an incorrect syllable sequence (Yoss & Darley, 1974).

The following characteristics were identified by direct assessment of the participant:

1. *Reduced diadochokinetic rate* (DDK) was indicated by a total function score on the Oral and Speech Motor Control Protocol (Robbins & Klee, 1987) of at least 1 *SD* below the normative mean for age, in conjunction with a normal structure score (administered to participants aged 6;11 [months;years] and younger at intake). Thirty-one of 33 participants (97%) who were administered this assessment had a total function score of at least 1 *SD* below the mean for their age. Two participants (ages 7;10 and 8;4) were administered the Oral and Speech Motor Control Protocol (Robbins & Klee, 1987) because of their inability to complete the Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate (hereinafter referred to as “Fletcher”; Fletcher, 1978); both scored over 3 *SDs* below the mean. The Fletcher was administered to participants 7 years of age and older. Reduced diadochokinetic rate was indicated by a score of at least 1 *SD* below the normative mean for age in the context of a normal oral peripheral examination. Three of the seven participants (43%) administered this assessment had DDK rates of at least 1 *SD* below the mean.
2. *Greater difficulty with polysyllabic words* was identified on the Multisyllabic Word Repetition (MSW) task (Catts, 1986) with 90% (36/40) of the participants obtaining a score of at least 2 *SDs* below the mean for age on this assessment (Lewis et al., 2004). A second measure composed of polysyllable non-words, the Nonsense Word Repetition (NWR) task (Kamhi & Catts, 1986), was administered, and a score of at least 2 *SDs* below the mean for age was identified for 85% (34/40) of the participants (Lewis et al., 2004). Additionally, the ability to pronounce one-, two-, and three-syllable words on the Goldman-Fristoe Test of Articulation (GFTA; Goldman & Fristoe, 1986) and the Goldman-Fristoe Test of Articulation–Second Edition (GFTA-2; Goldman & Fristoe, 2000) was noted to be better than multisyllable word and nonsense word production. Eighty-five percent of the participants with sCAS obtained standard scores on the GFTA or GFTA-2 that were

higher than their standard scores on both the MSW and NWR tasks at the initial assessment. Despite improvement in single word articulation on the GFTA and GFTA-2, this observation remained consistent at the time of follow-up (see Table 2 for scores for GFTA, MSW, and NWR).

3. *Restricted phonemic repertoire* was evidenced by deflated scores on the GFTA and GFTA-2 for younger participants (3–8 years of age; Goldman & Fristoe, 1986, 2000). Seventy-five percent of the participants (3–8 years of age) obtained a score on the GFTA or GFTA-2 that was at least 1.5 *SDs* below the mean for their age.
4. *Nondevelopmental phonological processes* were determined by the Khan–Lewis Phonological Analysis (both first and second editions; Khan & Lewis, 1986, 2002). Seventy-two percent of the younger participants (3–8 years of age) demonstrated at least one of the following nondevelopmental processes: glottal replacement, initial consonant deletion, and backing.

Inclusion Criteria for SSD-no sCAS

One hundred nineteen participants classified with SSD-no sCAS were selected from the larger longitudinal study sample as a comparison group for the current study. Individuals were independent (one per family). They met all the inclusion requirements for the larger SSD sample (see above for specific criteria for inclusion in the larger SSD study), had no history of a diagnosis of CAS, and did not meet the criteria stated above for sCAS. They were selected for comparison because they were in the same age range as the sCAS group (7–18 years). At intake, participants with SSD-no sCAS demonstrated a range of as few as two articulation errors to as many as 38 articulation errors on the GFTA or GFTA-2 (Goldman & Fristoe, 1986, 2000). Participants with SSD-no sCAS also demonstrated developmental phonological processes with usage ranging from “average” to “excessive,” as measured by the Khan–Lewis Phonological Analysis (Khan & Lewis, 1986) or the Khan–Lewis Phonological Analysis–Second Edition (Khan & Lewis, 2002).

Procedure and Measures

All participants were tested individually in two approximately 3-hr sessions by a licensed, ASHA-certified speech-language pathologist with experience in assessment of children with SSD for the initial assessment and all follow-up assessments. During these tests, frequent breaks were offered to reduce the effects of fatigue on test results. Testing was carried out in a speech research laboratory at Case Western Reserve University. At the parent’s request, testing was also carried out in a quiet and adequately lit room in the family’s home. Initial responses were transcribed while the child was speaking using broad phonetic transcription and also audio-recorded for later fine transcription. The institutional review board of University Hospitals

of Cleveland approved the study, and an informed consent/assent was obtained from all participants prior to testing.

The Word Attack and Word Identification subtests of the Woodcock Reading Mastery Tests–Revised (WRMT-R; Woodcock, 1987) were employed to assess single-word decoding skills and to create average-proficiency and low-proficiency reader groups. The Word Attack subtest provides a measure of phonics application for unknown words (nonsense words), and the Word Identification subtest provides a measure of sight vocabulary that can be read fluently and automatically. Average-proficiency readers were defined as children with standard scores of > 85 on both WRMT-R subtests, and low-proficiency readers were defined as those with standard scores of ≤ 85 on at least one of the WRMT-R subtests. Cutoff scores of 1–1.5 *SDs* below the normative mean or higher are often used by researchers and clinicians investigating specific language impairment (Records & Tomblin, 1994; Tomblin et al., 1997) and reading impairment (Flax et al., 2003).

The following measures were used to confirm the original diagnosis of sCAS (see Inclusion Criteria for sCAS section): MSW, NWR, Fletcher, GFTA, and GFTA-2; they were also used in conjunction with a Language Composite, an Elision task, Rapid Automatic Naming (RAN), and PIQ to compare sCAS and SSD-no sCAS groups at follow-up and as potential predictors of reading skills (see below).

The MSW (adapted from Catts, 1986) was used to assess speech production in real, complex words. This task has been used to evaluate the rapid selection and sequencing of phonemes and the execution of sequential motor commands. Participants were asked to repeat 20 multisyllable words presented by the examiner. The responses were recorded, phonetically transcribed, and analyzed for phonological processes and syllable structure. The percentage of words that were produced correctly was calculated and transformed into *z* scores. Scores considered in the analysis were age-corrected *z* scores based on the performance of unaffected siblings of probands ($n = 548$), assessed as part of the larger study.

The NWR (adapted from Kamhi & Catts, 1986) assessed phonological encoding and syllable sequencing abilities in polysyllabic nonsense words. Participants were asked to repeat 15 multisyllable nonsense words. The responses were recorded, phonetically transcribed, and analyzed for phonological processes and syllable structure. The percentage of words that were produced correctly was calculated and transformed into *z* scores for data analysis. Scores considered in analysis were age-corrected *z* scores based on the performance of unaffected siblings of probands ($n = 546$), assessed as part of the larger study.

Motor speech skills were measured using the Fletcher (Fletcher, 1978), which evaluates a child's ability to rapidly repeat single syllables (pΛ, tΛ, kΛ, fΛ, lΛ), bisyllables (pΛtə, pΛkə, tΛkə), and multisyllable sequences (pΛtəkə). Participants were asked to produce five monosyllabic repetitions, three 2-syllable repetitions, and one multisyllable repetition. Scores considered in the analysis were age-adjusted standard scores based on normative standards.

The Sounds-in-Words subtest of the GFTA and GFTA-2 (Goldman & Fristoe, 1986, 2000) was administered to assess a participant's articulation skill when labeling single words. Scores considered in the analysis were age-adjusted standard scores based on normative standards. The versions were merged by converting GFTA percentiles to standard scores compatible with GFTA-2.

A Language Composite was derived from one of the following overall language tests to assess expressive and receptive language skills: Clinical Evaluation of Language Fundamentals–Revised (Semel, Wiig, & Secord, 1987), Clinical Evaluation of Language Fundamentals–Third Edition (Semel, Wiig, & Secord, 1995), or Clinical Evaluation of Language Fundamentals–Fourth Edition Screening Test (Semel, Wiig, & Secord, 2003), and the Test of Language Development–Primary: Second Edition (Newcomer & Hammill, 1988) or the Test of Language Development–Primary: Third Edition (Newcomer & Hammill, 1997). Participants who enrolled in the study after the release of a newer version of a test battery were given the newer version. Scores considered in the analysis were age-adjusted standard scores based on normative standards.

Elision, a phonological manipulation task (e.g., say “stop” without /s/—“top”), was administered to assess phonological processing skills. The Elision subtest was taken from either the Comprehensive Tests of Phonological Processing–Experimental Version (Torgesen & Wagner, 1994) or the Comprehensive Test of Phonological Processing (Wagner, Torgesen, & Rashotte, 1999). Scores considered in the analysis were age-adjusted standard scores based on normative standards.

RAN; (Rapid Color Naming subtest) was administered to assess lexical retrieval and speed of processing of phonological information. The following normative data were used: Denckla and Rudel (1974), Comprehensive Tests of Phonological Processing–Experimental Version (1994), and Comprehensive Test of Phonological Processing (1999). *Z* scores based on the means and standard deviations of Denckla and Rudel or age-adjusted standard scores based on normative standards from the Comprehensive Test of Phonological Processing were used in the analysis.

PIQ, as measured by the Wechsler Intelligence Scale for Children–Third Edition (Wechsler, 1991), Wechsler Intelligence Scale for Children–Fourth Edition (Wechsler, 2003), Wechsler Adult Intelligence Scale–Revised (Wechsler, 1981), or Wechsler Adult Intelligence Scale–Fourth Edition (Wechsler, 2008), was administered to assess fluid reasoning, spatial processing, and visual–motor integration. Composite scores based on age-adjusted, normative standard scores were entered into data analysis.

Statistical Analysis

Associations of sCAS subset (low vs. average reading proficiency), with measures of oral language abilities, phonemic awareness, rapid automatic naming, diadochokinetic rates, single word articulation, and nonsense and multisyllabic word repetition, were determined by examining their

differences in distribution (Research Question 1). Associations of the low versus average reading proficiency subsets of the SSD-no sCAS group with these measures were similarly determined, as were associations of diagnostic category within low- and average-proficiency reading groups (sCAS average vs. SSD-no sCAS average proficiency, and sCAS low vs. SSD-no sCAS low proficiency; Research Question 2). Associations of the speech and language measures with reading level and diagnostic category were tested for statistically significant differences using the parametric Welch's two-sample *t* test or the nonparametric Wilcoxon–Mann–Whitney two-sample exact test, depending on appropriateness. Estimates of median differences and confidence intervals were calculated using the Hodges–Lehmann estimator, which is consistent with the Wilcoxon test. Statistical significance was determined at the .05 level, with adjustments for multiple testing applied and noted accordingly.

The best predictors of average-proficiency reading in the combined sCAS and SSD-no sCAS groups (Research Question 3) were identified using multivariate logistic regression. The regression model including the best predictors was determined via backward stepwise model selection. At each step, the Wald test χ^2 statistic was computed, which compared the current model to the preceding model. Variable removal at each step was determined by the largest *p* value. The most parsimonious, best fitting model was determined to be the one that was a significantly better fit (Wald $p < .05$) than the null model with the fewest predictors (i.e., additional predictors did not offer a significant improvement in fit). For all statistical techniques used, assumptions regarding the data were checked and satisfied.

Results

Comparisons Across Reading Proficiency Groups on Demographic and Reading Measures

Of the 40 children with sCAS, 26 (65%) were low-proficiency readers, while only 29 (24%) of the 119 with SSD-no sCAS were low-proficiency readers. The percentage of low-proficiency readers within the sCAS group was significantly higher than those in the SSD-no sCAS group, $\chi^2(1) = 20.1, p < .001$. Comparisons across diagnostic reading groups (sCAS average vs. low proficiency and SSD-no sCAS average vs. low proficiency) failed to reveal significant differences in age, gender, or socioeconomic status. However, average-proficiency readers with sCAS and SSD-no sCAS had higher mean and median scores compared to low-proficiency readers with sCAS and SSD-no sCAS for PIQ, Word Identification, and Word Attack (see Table 1).

Skill Differences Between Reading Proficiency Groups for Participants With sCAS

Table 2 presents results for skill differences between reading proficiency groups for children with sCAS. The

Bonferroni adjusted alpha level for significant differences in distributions was .007. Low-proficiency readers with sCAS obtained significantly lower scores than the average-proficiency readers with sCAS on Elision ($Z = 3.6, p < .001$), Language Composite ($Z = 3.9, p < .001$), NWR ($Z = 3.0, p = .003$), and Fletcher ($Z = 3.5, p < .001$). No significant differences were demonstrated for RAN ($Z = 1.5, p = .13$), MSW ($Z = 2.0, p = .045$), or GFTA ($Z = 0.7, p = .48$; see Table 2).

Table 3 presents results for skill differences between reading proficiency groups for children with SSD-no sCAS. The Bonferroni adjusted alpha level for significant differences was .007. Low-proficiency readers with SSD-no sCAS obtained significantly lower scores than the average-proficiency readers with SSD-no sCAS on Elision ($t = 5.9, p < .001$), Language Composite ($t = 7.6, p < .001$), NWR ($t = 3.9, p < .001$), and MSW ($Z = 4.0, p < .001$). No significant differences were demonstrated for RAN ($t = 2.8, p = .009$), Fletcher ($t = 1.1, p = .29$), or GFTA ($Z = -2.0, p = .04$; see Table 3).

Skill Differences Within Reading Proficiency Groups Across Diagnostic Category (Average-Proficiency Readers With sCAS vs. Average-Proficiency Readers With SSD-no sCAS and Low-Proficiency Readers With sCAS vs. Low-Proficiency Readers With SSD-no sCAS)

Table 4 presents results for skill differences between diagnostic groups with the same level of reading proficiency (average and low proficiency). The level of significance after applying the Bonferroni multiple testing correction was .0056. The subset of the sCAS group with average-proficiency reading had significantly lower scores than the SSD-no sCAS group with average-proficiency reading, significantly differing in its score distribution on NWR ($Z = 2.7, p = .005$) and Fletcher ($Z = 3.4, p < .001$); analysis failed to reveal, however, significant differences for Word Identification, Word Attack, Language Composite, RAN, Elision, MSW, and GFTA. Similarly, the low-proficiency readers from the sCAS group had significantly lower scores than the low-proficiency readers from the SSD-no sCAS group on NWR, $t(51.1) = 3.4, p = .001$, Fletcher ($Z = 8.2, p < .001$), and GFTA ($Z = 4.2, p < .001$), whereas no significant differences were found for Word Identification, Word Attack, Language Composite, RAN, Elision, and MSW.

Multivariate Predictors of Average-Proficiency Reading in sCAS and SSD-no sCAS

Results from multivariate logistic regression to identify the best predictors of average-proficiency reading in children with SSD-no sCAS and sCAS are presented in Table 5. Initial predictors in the model included group (sCAS vs. SSD-no sCAS), age, PIQ, and the speech and language measures (Language Composite, Elision, MSW, NWR, GFTA, RAN, and Fletcher; see Figure 1 for a correlation

Table 1. Demographics for the suspected childhood apraxia of speech (sCAS) and speech sound disorder but no sCAS (SSD-No sCAS) reading groups.

Variable	Summary measures	sCAS		SSD-No sCAS		Comparison across all diagnostic reading groups
		Average-proficiency readers	Low-proficiency readers	Average-proficiency readers	Low-proficiency readers	
		<i>n</i> = 14	<i>n</i> = 26	<i>n</i> = 90	<i>n</i> = 29	
Age	<i>M</i> (<i>SD</i>) [<i>Mdn</i>] Range	11.1 (3) [10.5] (7.25–16.16)	11.9 (2.8) [11.1] (8.25–18)	12.5 (3) [12.5] (7.4–17.9)	11.6 (3) [11.2] (7.3–17.8)	.19 ^a
Sex	Female (%)	3 (21.4%)	5 (19.2%)	33 (36.7%)	8 (27.6%)	.31 ^b
	Male (%)	11 (78.6%)	21 (80.8%)	57 (63.3%)	21 (72.4%)	
PIQ	<i>M</i> (<i>SD</i>) [<i>Mdn</i>] Range	102.0 (14.4) [101] (84–136)	92.3 (17.8) [90] (63–129)	110.3 (12.9) [111] (66–136)	96.2 (15) [96] (60–120)	< .001 ^a
	Word Attack	102.4 (7.8) [100.5] (93–125)	76.7 (10.9) [78.5] (57–94)	101.6 (9.0) [101] (86–122)	71.9 (14.1) [76] (33–94)	
Word ID	<i>M</i> (<i>SD</i>) [<i>Mdn</i>] Range	99.1 (9.1) [97] (89–119)	76.2 (9.9) [78.5] (56–99)	102.9 (9.7) [101] (88–143)	73.5 (14.2) [78] (45–102)	< .001 ^c
	SES	1 2 3 4 5	0 1 7 11 7	3 6 13 39 29	3 4 7 8 7	

Note. Average-proficiency readers' WRMT-R Word Attack and Word Identification standard scores were above 85; low-proficiency readers' WRMT-R Word Attack or Word Identification standard scores were at or below 85. Socioeconomic categories were based on the Hollingshead Four-Factor Index of Social Status (Hollingshead, 1975) with 1 = lowest SES and 5 = highest SES. PIQ = performance IQ; WRMT-R = Woodcock Reading Mastery Tests–Revised; SES = socioeconomic status.

^aKruskal–Wallis test. ^bFisher exact test. ^cAnalysis of variance.

among the test measures). The only significant predictors of low reading proficiency in the final, reduced model were Language Composite, Elision, MSW, and Fletcher. The likelihood of being an average-proficiency reader (vs. low-proficiency reader) increases by 11% ($OR = 1.109$, $p < .001$, 95% CI [1.06, 1.17]) with each 1-point increase in standard score on Language Composite, by 7% ($OR = 1.073$, $p < .001$, 95% CI [1.04, 1.12]) with each 1-point increase in standard score on Elision, by 4% ($OR = 1.036$, $p = .008$, 95% CI [1.01,

1.07]) with each 1-point increase in standard score on MSW, and by 4% ($OR = 1.073$, $p = .03$, 95% CI = [1.01, 1.08]) with each 1-point increase in standard score on Fletcher.

Discussion

The association of literacy problems with CAS has been a topic of debate, with some researchers viewing these

Table 2. Comparisons of average- and low-proficiency readers on speech and language measures in the suspected childhood apraxia of speech (sCAS) group.

Variable	sCAS group								Test statistic	p
	Average-proficiency readers				Low-proficiency readers					
	n = 14				n = 26					
	M	SD	Mdn	Range	M	SD	Mdn	Range		
RAN	99.3	13.6	100.0	(80–125)	89.2	19.1	92.5	(55–120)	1.5 ^a	.13
Elision	103.6	17.0	100.0	(80–150)	81.5	15.4	80.0	(55–115)	3.6 ^a	< .001*
LC	98.9	13.8	97.0	(80–131)	76.8	14.7	79.0	(50–97)	3.9 ^a	< .001*
NWR	82.5	11.6	82.5	(59–99)	67.5	16.1	64.8	(47–112)	3.0 ^a	.003*
MSW	76.1	23.8	75.5	(39–109)	60.8	18.6	58.8	(31–98)	2.0 ^a	.045
Fletcher	92.6	10.3	95.0	(70–111)	75.2	14.7	75.0	(55–100)	3.5 ^a	< .001*
GFTA	93.3	12.6	94.5	(73–108)	91.3	20.7	90.5	(61–135)	0.7 ^a	.48

Note. *p* values shown are unadjusted. The Bonferroni adjusted alpha (α) significance level was .007 (familywise $\alpha = .05$). Significant differences after multiple testing correction are indicated with an asterisk (*). RAN = Rapid Automatic Naming; LC = Language Composite; NWR = Nonsense Word Repetition Task; MSW = Multisyllabic Word Repetition Task; Fletcher = Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate; GFTA = Goldman–Fristoe Test of Articulation.

^aWilcoxon–Mann–Whitney two-sample exact test was used.

Table 3. Comparisons of average- and low-proficiency readers on speech and language measures in the speech sound disorder but no suspected childhood apraxia of speech (SSD-no sCAS) group.

SSD-no sCAS group										
Variable	Average-proficiency readers				Low-proficiency readers				Test statistic	p
	n = 90				n = 29					
	M	SD	Mdn	Range	M	SD	Mdn	Range		
RAN	101.8	14.6	100.0	(75–150)	91.9	17.4	90.0	49–134	2.8 ^a	.009
Elision	103.8	11.0	105.0	(60–125)	83.8	17.0	81.0	55–143	5.9 ^a	< .001*
LC	101.3	14.6	102.0	(56–139)	77.8	14.5	81.0	50–102	7.6 ^a	< .001*
NWR	94.3	15.3	95.1	(54–122)	81.9	14.8	83.7	54–102	3.9 ^a	< .001*
MSW	91.4	23.5	97.0	(37–122)	70.5	21.5	70.0	33–121	4.0 ^b	< .001*
Fletcher	106.7	14.4	103.2	(83–147)	103.9	11.6	102.1	85–128	1.1 ^a	.29
GFTA	105.3	19.2	103.0	(46–135)	115.7	18.2	108.7	88–135	–2.0 ^b	.04

Note. p values shown are unadjusted. The Bonferroni adjusted alpha (α) significance level was .007 (familywise α = .05). Significant differences after multiple testing correction are indicated with an asterisk (*). RAN = Rapid Automatic Naming; LC = Language Composite; NWR = Nonsense Word Repetition Task; MSW = Multisyllabic Word Repetition Task; Fletcher = Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate; GFTA = Goldman-Fristoe Test of Articulation.

^aWelch's two-sample t test. ^bWilcoxon–Mann–Whitney two-sample exact test.

problems as present in a majority of children with CAS (Lewis et al., 2004; Nijland et al., 2015; Orton, 1964; Peter et al., 2013, 2016; Worthey et al., 2013) and others conceptualizing poor literacy as merely one of a collection of traits that can co-occur with CAS (ASHA, 2007, n.d.; Hall et al., 2006). However, few studies have explicitly focused on rates and correlates of reading problems in children with CAS. In the current study, we compared average-proficiency readers with sCAS to low-proficiency readers with sCAS on motor speech difficulties associated with CAS and on multiple cognitive–linguistic skills with established associations with reading skills (i.e., oral language abilities, phonological awareness, and RAN). A comparison group of students with histories of SSD-no sCAS was employed to determine

if the deficits observed in low-proficiency readers with sCAS were similar to those observed in individuals with other forms of SSD. Our findings indicate that students with sCAS and low reading proficiency may exhibit linguistic skill deficits in common with students with other forms of SSD with low-proficiency decoding skills. Furthermore, the association between difficulty in more complex articulation and motor speech tasks (i.e., MSW and DDK rate) and reading proficiency in the sCAS and SSD-no sCAS groups suggests that weaknesses in these areas signal additional risk for decoding problems in the broader spectrum of children with SSD.

Our first research aim was to examine differences between children with sCAS and average reading proficiency

Table 4. Comparisons of difference scores between the suspected childhood apraxia of speech (sCAS) and speech sound disorder but no sCAS (SSD-no sCAS) groups on reading and speech-language measures within reading proficiency groups.

Measure	Average-proficiency readers			Low-proficiency readers		
	Difference [95% CI] ^a	Test statistic	p	Difference [95% CI] ^a	Test statistic	p
Word ID	3.7 [–1.8, 9.3] ^b	1.4 ^b	.17	–2.7 [–9.3, 3.8] ^b	–0.8 ^b	.41
Word Attack	–0.9 [–5.7, 3.9] ^b	–0.4 ^b	.71	–4.7 [–11.5, 2.1] ^b	–1.4 ^b	.17
LC	4 [–4, 11] ^c	0.9 ^c	.35	1 [–8, 9] ^b	0.2 ^c	.84
RAN	1.4 [–5, 10] ^c	0.4 ^c	.66	0.2 [–9.2, 11.2] ^c	0.1 ^c	.90
Elision	5 [–3, 10] ^c	0.8 ^c	.41	2.3 [–6.5, 11.0] ^b	0.5 ^b	.61
MSW	14.1 [2.9, 30.3] ^c	2.3 ^c	.02	9.4 [–2.4, 19.8] ^c	1.6 ^c	.12
NWR	12.5 [3.9, 20.7] ^c	2.7 ^c	.005*	14.4 [6.0, 22.8] ^b	3.4 ^b	.001*
Fletcher	12.5 [4.6, 20.6] ^c	3.4 ^c	< .001*	28.2 [20.8, 36.8] ^c	8.2 ^c	< .001*
GFTA	13.4 [–0.6, 22] ^c	1.7 ^c	.09	27.5 [12, 38] ^c	4.2 ^c	< .001*

Note. p values and confidence intervals shown are unadjusted. The Bonferroni adjusted alpha (α) significance level was .0056 (familywise α = .05). Differences significant after multiple testing correction are indicated with an asterisk (*). LC = Language Composite; RAN = Rapid Automatic Naming; MSW = Multisyllabic Word Repetition Task; NWR = Nonsense Word Repetition Task; Fletcher = Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate; GFTA = Goldman-Fristoe Test of Articulation.

^aDifference (SSD-no sCAS vs. sCAS) shown is mean difference for the Welch's two-sample t test and median difference, calculated using the Hodges–Lehmann estimator for the Wilcoxon–Mann–Whitney test. ^bWelch's two-sample t test was used. ^cWilcoxon–Mann–Whitney two-sample exact test.

Table 5. Model selection results for the combined suspected childhood apraxia of speech (sCAS) and speech sound disorder but no sCAS groups.

Variable	Model 1	Model 2	Model 3	Model 4	Model 5	Model 6	Model 7
LC	1.10** (0.03)	1.10** (0.03)	1.10** (0.03)	1.10** (0.03)	1.10** (0.03)	1.11** (0.03)	1.11** (0.03)
Elision	1.07** (0.02)	1.07** (0.02)	1.07** (0.02)	1.07** (0.02)	1.07** (0.02)	1.08** (0.02)	1.07** (0.02)
MSW	1.05* (0.02)	1.05* (0.02)	1.05* (0.02)	1.06** (0.02)	1.05** (0.02)	1.04** (0.01)	1.04** (0.01)
Fletcher	1.04 (0.03)	1.05* (0.02)	1.05* (0.02)	1.05* (0.02)	1.04 (0.02)	1.04 (0.02)	1.04* (0.02)
RAN	1.04 (0.02)	1.04 (0.02)	1.04 (0.02)	1.04 (0.02)	1.04 (0.02)	1.03 (0.02)	
GFTA	0.97* (0.02)	0.97* (0.02)	0.97* (0.02)	0.97* (0.01)	0.98 (0.01)		
Age	0.85 (0.12)	0.85 (0.12)	0.85 (0.12)	0.85 (0.11)			
NWR	1.02 (0.02)	1.02 (0.02)	1.02 (0.02)				
PIQ	1.00 (0.02)	1.00 (0.02)					
sCAS	0.93 (0.87)						
$\chi^2_{(df = 1)}$		0.008 ($p = .93$)	0.05 ($p = .82$)	0.92 ($p = .34$)	2.20 ($p = .14$)	2.63 ($p = .10$)	2.87 ($p = .09$)

Note. Backward stepwise model selection was implemented. The variable with the largest p value was removed at each step until a significant difference in model fit was reached. The χ^2 test statistic shown is for the Wald test (comparing the fit of the model with one predictor removed to the fit of the previous model). The estimates shown are e^B = exponential B , which can be interpreted as odds ratio, and in parentheses are the standard error of B . LC = Language Composite; MSW = Multisyllabic Word Repetition Task; Fletcher = Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate; RAN = Rapid Automatic Naming; GFTA = Goldman-Fristoe Test of Articulation; NWR = Nonsense Word Repetition Task; PIQ = performance IQ.

* $p < .05$. ** $p < .01$.

and children with sCAS and low-proficiency reading on measures of speech- and language-related skills. A high rate of low-proficiency readers with sCAS (26/40, 65%) compared to average-proficiency readers with sCAS (14/40, 35%) was observed in our sample. This higher percentage of low-proficiency readers with sCAS is less consistent

Figure 1. Correlation matrix for included test measures. Lighter colors represent a stronger correlation. RAN = Rapid Automatic Naming; PIQ = performance IQ; LC = Language Composite; GFTA = Goldman-Fristoe Test of Articulation; Fletcher = Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate; MSW = Multisyllabic Word Repetition Task; NWR = Nonsense Word Repetition Task.

with the conceptualization of CAS as strictly a motor speech deficit than with one that considers reading disorder a part of the spectrum of disabilities associated with CAS. Reading disorders in the general population are substantially lower than in the present cohort of children with sCAS, as estimates of the prevalence of reading disabilities in children in the general population range from 5% (Moats & Dakin, 2008) to between 5% and 17% (Shaywitz et al., 1998).

However, not all participants with sCAS demonstrated low-proficiency reading. Differences in performance on speech and language measures between the average- and low-proficiency readers with sCAS identified specific risk factors for literacy difficulties. The oral language and phonological awareness abilities that have been typically associated with the development of decoding were intact for average-proficiency readers with sCAS, but not for low-proficiency readers. Average-proficiency readers also performed better on the RAN than low-proficiency readers, although group differences on the RAN did not reach statistical significance. Other researchers have described subgroups of children with CAS and accompanying language impairment (Ekelman & Aram, 1983; Lewis et al., 2004; Thoonen et al., 1997; Zuk, Iuzzini-Seigel, Cabbage, Green, & Hogan, 2018). For example, Zuk et al. (2018) found evidence of deficits in speech perception in a subgroup of children with CAS with accompanying language impairment. Speech perception deficits have also been linked to phonological representations required for accurate decoding (Brady, Shankweiler, & Mann, 1983; Lieberman, Meskill, Chatillon, & Schupack, 1985; McBride-Chang, 1996). Decoding deficits in a subset of the present group of children with sCAS, therefore, may be partially explained by endophenotypes, such as oral language, phonological awareness, and speech perception deficits.

The subset of children with sCAS and average-proficiency reading also performed significantly better than children with sCAS and low-proficiency reading on tests of motor speech sequencing abilities (DDK rates) and NWR, although both groups obtained scores that were well below the normative mean on NWR. NWR is hypothesized to measure a variety of skills including phonological encoding and phonological short-term memory demonstrated to be important for the development of decoding skills (Graf Estes, Evans, & Else-Quest, 2007). NWR also measures oral motor praxis, motor speech sequencing, and articulation (Krishnan et al., 2017; Reuterskiöld & Grigos, 2015); deficits in these skills can confound the assessment of NWR in children with motor speech disorders (Coady & Evans, 2008; Graf Estes et al., 2007; Krishnan et al., 2017; Shriberg et al., 2009). It is possible that deficits in NWR reflect a greater contribution of motor speech ability for the sCAS average-proficiency reading group, while the sCAS low-proficiency reading group's more extensive speech and language problems reflect a contribution of deficits in both motor speech and phonological processing skills.

Consistent with the predictive validity of MSW in identifying CAS (Murray, McCabe, Heard, & Ballard, 2015), both sCAS reading groups demonstrated difficulty on this measure. Similar to NWR, MSW places demands on several underlying skills, including phonological encoding, phonological memory, transcoding (formulating a motor plan), and speech articulation. Unlike NWR, however, MSW may be more directly associated with exposure to real words that contain complex phonological sequences. Previous studies have revealed that children with sCAS, as well as those with less severe forms of SSD, have persisting weaknesses in producing multisyllable words (Lewis et al., 2015; Peter, Lancaster, Vose, Middleton, & Stoel-Gammon, 2018; Peter et al., 2016; Preston & Edwards, 2007; Preston et al., 2014). Deficits in phonological encoding, phonological memory, and motor planning and programming associated with MSW can persist into adulthood for some individuals with CAS and affect reading and spelling (Peter et al., 2013, 2018). Lewis et al. (2015) found that adolescents with either a resolved or persistent SSD who evidenced difficulty on an MSW task demonstrated deficits in single word decoding.

The second research aim was to examine differences between the two diagnostic groups, sCAS and SSD-no sCAS, at each level of reading proficiency (low vs. average). Findings failed to reveal differences between the low-proficiency subsets of these two groups on measures of oral language and phonological awareness (Elision) and RAN, with both subsets scoring well below average on the oral language and phonological awareness measures and within the low-average range on the RAN. Comparisons of the average-proficiency subsets of the two groups also failed to reveal significant differences on these measures, with mean scores for these measures falling solidly within the average range for both subsets of children. Not all children with SSD, including those with sCAS, have accompanying language difficulties and consequent reading problems, thus demonstrating heterogeneity within the sCAS group possibly due

to genetics, environment, or intervention (Lewis et al., 2004, 2015). While the contribution of cognitive-linguistic skills to decoding has been well established for other forms of SSD, the current study documented that these same abilities distinguish good and poor decoding in children with sCAS.

As expected, significant differences were observed between SSD-no sCAS and sCAS average- and low-proficiency reading groups on measures that assess motor speech sequencing abilities considered to be characteristic of CAS (i.e., DDK rate, Fletcher). Both reading proficiency subsets of the sCAS groups performed more poorly than the corresponding subsets of the SSD-no sCAS groups on this measure. Differences between the sCAS and SSD-no sCAS groups were also observed on an NWR, with readers with sCAS scoring lower than readers with SSD-no sCAS regardless of reading proficiency. Difficulties with multisyllable repetition tasks, such as NWR, reflect a complex relationship between motor speech ability and cognitive-linguistic processing requirements (Reuterskiöld & Grigos, 2015), especially for children with motor speech deficits. Difficulties that the SSD-no sCAS low-proficiency reading group demonstrated on both MSW and NWR in comparison to the average-proficiency readers with SSD-no sCAS likely reflect greater challenges with cognitive-linguistic processing that have been traditionally associated with a core phonological deficit in this subtype of SSD (Anthony et al., 2011; Cabbage et al., 2018; Catts, 1986; Pennington & Bishop, 2009; Rvachew & Brosseau-Lapre, 2018).

Our third research aim was to identify the best predictors of decoding skills in the sCAS and SSD-no sCAS groups. As expected, oral language and phonological awareness skills best predicted decoding proficiency in both groups. These findings are consistent with evidence that children with reading impairment suffer from a core deficit in phonological processing (Ramus & Szenkovits, 2008; Share & Stanovich, 1995; Snowling, 1998; Wagner & Torgesen, 1987) and broad-based linguistic deficits, such as accompanying oral language disorders (Bishop & Adams, 1990; Kamhi & Catts, 2012; Lombardino, Riccio, Hynd, & Pinheiro, 1997). In a longitudinal study of 183 second-grade children diagnosed as reading disabled, Catts, Fey, Zhang, and Tomblin (1999) found that oral language (i.e., a composite score of expressive and receptive abilities) made a unique contribution to single word decoding above and beyond that of phonological awareness.

Two additional measures related to speech production predicted decoding, the MSW and DDK tasks. This finding suggests that speech production also contributes to decoding abilities in some children with SSD. As outlined above, MSW measures a complex subset of skills including phonological encoding, phonological short-term memory, and transcoding (translating a phonological plan into a motor plan). Phonological encoding and phonological short-term memory skills have been linked directly to the establishment of accurate phonological representations required for fluent decoding skills hypothesized to be deficient in SSDs with a cognitive-linguistic basis. These tasks also place demands on transcoding and speech production, making it difficult

to evaluate their unique contributions to decoding for children with CAS.

To isolate speech production demands from the cognitive-linguistic and memory demands in MSW, some researchers have examined the association between DDK, a simpler productive syllable sequencing task, and decoding. Fawcett and Nicolson (2002) demonstrated that two groups of dyslexic adolescents ($M_{\text{age}} = 13$ and 16 years), with no overt speech errors, exhibited decreased rates of articulation and prolonged pauses between repetitions on DDK tasks relative to the normally achieving controls. The authors proposed that the pauses were indicative of slower access to phonological representations and motor planning difficulties and that the observed reduced rate of articulation was indicative of slower production of articulatory gestures (Fawcett & Nicolson, 2002). Arthur, Preston, and Pugh (2010) examined DDK rate (speed in single syllable production) and accuracy (accurate alternating syllable production) and its relationship to word reading proficiency along with a variety of cognitive-linguistic assessments for 78 child participants between the ages of 7 and 12 years. Accuracy in the alternating syllable sequencing task accounted for a small but unique amount of variance in decoding skill beyond that of IQ, phonological awareness (Elision), and age.

It is not surprising then that MSW and DDK surfaced as significant contributors to reading accuracy in the current study. At the time of assessment, all the participants had a history or an existing diagnosis of speech impairment. Estimates indicate that roughly 25% of children with reading disability have a history of “clinically significant speech production” difficulties in early childhood (Pennington & Leffly, 2001, p. 830) and that 30% of children with a history of SSD are diagnosed with reading disability at school age (Lewis, 1996). Scores of single word articulation on the GFTA and the diagnosis of sCAS also entered in the model were not predictive of reading group assignment. It is likely that the complex speech production demands of MSW and the motor speech sequencing demands of DDK better captured abilities such as phonological encoding, phonological access, and speed of articulatory gestures, associated with decoding than single word articulation at school age. Furthermore, because the DDK task was one of the tests used to classify children into the sCAS group, the association of this measure with reading proficiency might have been due in part to the number of low-proficiency sCAS readers with pronounced difficulties in motor speech sequencing abilities.

Clinical Implications

The ability domains that best distinguished children with sCAS with average-proficiency compared to low-proficiency reading skill were oral language, phonological awareness, and speech production, areas in which speech and language clinicians have clinical expertise. ASHA has placed literacy intervention within the scope of practice for speech-language pathologists and states that speech and

language clinicians play a pivotal role in the development of literacy for children and adolescents who have both speech-language and written language impairment (ASHA, 2001, 2016). The implication of these findings for speech and language clinicians is that an early childhood diagnosis of CAS is often accompanied by literacy impairment at school age. Similar to other children with speech and language deficits at risk for decoding problems, it would be preferable to design speech and language therapy for children with CAS that addresses the intersection of speech, language, and literacy deficits early on. Appropriate literacy intervention would include systematic phonological awareness intervention coupled with age-appropriate, explicit, and systematic sound-symbol relationship training and morphological awareness instruction (ASHA, 2016; Bowers, Kirby, & Deacon, 2010).

A proposed intervention for children with CAS utilizing an “integrated phonological approach” combining speech, phonological awareness, and sound-symbol associations yielded positive gains in speech proficiency, phonological awareness, and decoding (Gillon & Moriarty, 2007; McNeill et al., 2009). A systematic review of treatment outcomes for children with CAS concluded that the “integrated phonological approach” showed large generalization effects for improved speech production (Murray et al., 2014). Additionally, it may be appropriate when assessing decoding skills in children with SSD, including those with sCAS, to utilize silent, word- and sentence-level decoding measures to eliminate the confounding effect of speech production difficulties when reading orally.

Limitations and Future Directions

A limitation of the current study was the lack of information on interventions for reading and speech-language therapy for study participants. Controlling for these interventions may have helped to account for differences in children’s reading and speech-language outcomes. An additional limitation was that the sole administered assessments of reading were measures of single and nonsense word decoding. For children with significant speech intelligibility deficits as in sCAS, tests that measure silent word reading or silent sentence reading might be better measures to disentangle decoding from difficulties with speech production. Future research is also needed to examine the nature and predictors of reading achievement in children with CAS using more comprehensive measures of phonological processing abilities, reading fluency, and reading comprehension. Longitudinal studies are likewise needed to chart the developmental trajectory of the disorder and examine reading outcomes for children with persistent versus recovered CAS. Future research comparing treatment paradigms such as an “integrated phonological approach” (Gillon & Moriarty, 2007; McNeill et al., 2009) with more traditional motor-based approaches would also be useful in determining the types of interventions that are most effective in improving literacy outcomes for this population.

Conclusion

The current conceptualization of CAS as primarily a motor speech disorder may encourage interventions that rely largely on habituating accurate motor speech plans with little attention paid to the development of literacy skills. Viewing CAS as a motor speech disorder that commonly includes reading impairment may be more useful to clinicians given the high percentage of children with CAS who exhibit decoding difficulties. Early literacy intervention for these children is crucial given the persistence of the speech and language disorder coupled with the pernicious effect of decoding difficulties on lifelong achievement (Nordström, Jacobson, & Söderburg, 2016; Stanovich, 1986). Zaretsky et al. (2010) suggest that literacy instruction for all children with a diagnosis of CAS needs to begin as early as preschool to shore up weaker phonological processing skills that can undermine decoding ability in this population.

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