THE ASSOCIATION BETWEEN AN EARLY DIAGNOSIS OF CHILDHOOD APRAXIA OF SPEECH AND WORD-LEVEL DECODING SKILLS

by

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DEDICATION

First, I wish to dedicate this dissertation research to my advisor, Dr. Barbara Lewis, whose research in speech, language, and literacy has been seminal. Her expertise, kindness, patience, and encouragement enabled me to start, stay focused, and finish my Ph.D. program successfully. How does one say thank you to such an amazing mentor except to promise to pay it forward in some capacity?

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ABSTRACT

The Association Between an Early Diagnosis of Childhood Apraxia
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Childhood Apraxia of Speech (CAS) is a developmental speech sound disorder (SSD) notable for its severity and persistence of speech difficulties. Debate has surfaced historically about the cause of the speech and language deficits observed in the disorder. As children with CAS often present with comorbid language problems (i.e., receptive and expressive), learning disabilities (i.e., reading and spelling), and fine and gross motor difficulties, any conceptualization of the disorder should include an accounting of the multiple domains affected. However, CAS has been primarily viewed as a motor-speech disorder affecting speech production with consequences for expressive language. While there is extensive research about the association between an early diagnosis of other idiopathic SSD and poorer literacy outcomes, there is limited research investigating this same association for CAS. Given the increased risk of reading disorder (RD) with other idiopathic SSD, a better understanding of this association for CAS is crucial for children with this diagnosis.

This dissertation research investigated literacy and literacy-related skills in a group of children with an early diagnosis of CAS (n = 16), ranging in age from 8 to 14 years. Comparisons were made with a group of children in a similar age range with a diagnosis of reading disorder without any history of SSD (RD-no SSD, n = 16). Results suggest that many children with an early diagnosis of CAS share the same degree of difficulty with word-level decoding as those diagnosed with RD-no SSD. In contrast, the two groups' phonological processing abilities were significantly different. The CAS group scored below the normative mean for phonological awareness and phonological memory, and the RD-no SSD group obtained mean scores within the average range.

Additionally, subgroups were identified within the CAS participant group by average and below-average word-level decoding fluency. Below-average decoding fluency was associated with persistent speech sound errors, and phonological awareness and phonological memory deficits. Overall, these findings suggest that individuals with an early diagnosis of CAS and individuals with RD-no SSD demonstrate similar impairments in reading skills. However, the endophenotypes that underlie these literacy difficulties may differ between the groups, requiring remediation tailored to meet specific needs.

Chapter 1. Theoretical Foundation for the Associations between CAS and Word-Level Decoding Skills

1.1 Models of SSD

Nearly 1 in 12 U.S. children between the ages of 3 and 17 have experienced a communication disorder within the last year (Black et al., 2015). Speech sound disorder (SSD), a "theory-neutral cover term" for childhood speech disorders (Shriberg, 2010, p. 2), is by far the most common form of childhood communication disorder. SSD affects nearly 42% of school-age children ages 3 to 10 and 24% of school-age children and adolescents ages 11 through 17 (Black et al., 2015). Difficulties with speech sound production interfere with communication and have consequences for affected children's social and emotional development when they cannot make themselves understood during peer and adult interactions (Mcleod et al., 2013). Children with SSD are also at risk for educational issues. Written language (i.e., reading and spelling) has as its foundation oral language abilities, specifically phonological processing. Phonological processing principally begins with encoding phonemes (i.e., speech sounds) of the ambient language which are used for spoken and written language (Wagner & Torgesen, 1987). Children with a history of SSD have an increased risk of phonological processing difficulties (Rvachew, 2007; Rvachew & Grawburg, 2006) and, therefore, a substantially increased risk for literacy difficulties (Nathan et al., 2004).

However, there are subtypes of SSD that can be distinguished by differences in the putative underlying cause (e.g., motor versus cognitive-linguistic) or the characteristics of the speech output (consistent error patterns versus errors related to motor planning). Furthermore,

different subtypes are proposed to vary in their impact on language and literacy. At least three models have been offered to describe the differing subtypes: the descriptive-linguistic model, the medical model, and the psycholinguistic framework (Namasivayam et al., 2020; Rvachew & Brosseau-Lapré, 2018; Shriberg et al., 2010; Stackhouse & Wells, 1997; Waring & Knight, 2013).

Dodd's Differential Diagnosis system (2005) is an example of the descriptive-linguistic model. The model classifies SSD subtypes based on "surface-level pattern errors that reflect underlying *subgroup-specific processing deficits*" (Waring & Knight, 2013, p. 31, emphasis author's). Dodd proposes three categories in her classification system: phonetic (articulation disorder), cognitive-linguistic (phonemic/phonological disorder), and motor-based speech disorder. Motor-based speech disorders include Childhood Apraxia of Speech (CAS), a motor planning and/or programming disorder, and Dysarthria, an execution or neuromuscular disturbance of speech. As described above, Dodd's surface-level patterns are proposed to link to different processing deficits (Dodd, 1995). For example, Broomfield and Dodd (2004) evaluated comorbidities for 320 children referred to a clinic with a primary speech delay. The authors suggested that "poor response" by the participants on an assessment of phonological awareness indicated a "theoretical *linguistic* basis" for some of the surface level patterns (p. 147, emphasis author's). A CAS diagnosis is based on a "cluster of symptoms that give rise to deviant speech production" (Broomfield & Dodd, p. 138), such as difficulty with phonetic assembly, oromotor difficulties, but intact phonological awareness skills.

The Speech Disorders Classification System (SDCS, Shriberg, 1993) is based on etiology, a *medical model* of classification (Shriberg et al., 2010). It was designed to classify SSD and conduct research into the genetics of SSD (Shriberg, 1993), especially SSD of unknown origin. Estimates as high as 98% of children with a diagnosis of SSD have an idiopathic form of the disorder (Shriberg, 2010). Shriberg et al. (2019) proposed three categories of speech disorders: Speech Delay, Speech Errors, and Motor Speech Disorders. Each of the three categories is presumed to have a distal cause (i.e., genomic, and environmental) and a proximal cause (i.e., auditory, somatosensory, planning and programming, or execution deficits). For the Speech Delay category, the core deficit is hypothesized to be "developmental delays in encoding and retrieving linguistic representations" (Shriberg et al., 2019, p. 680).

The psycholinguistic model (Stackhouse & Wells, 1997) is a framework that details specific levels of breakdown in a child's speech processing system. The model proposes three main levels: an input processing level, a representational level (i.e., stored lexical knowledge), and a speech output level. A child can hypothetically have difficulty with one or all three levels, and difficulties at lower levels of input processing can impact ascendant levels. For example, disruptions at the input level, such as phonetic discrimination and phonological recognition can impede accurate storage of phonological representations, derailing a child's ability to form robust phoneme representations required for both speech production and written language development.

All three theoretical models reviewed above include CAS as a subtype of SSD. The descriptive-linguistic and psycholinguistic models propose that CAS is a multi-deficit disorder

involving at least three stages: the creation of a phonological plan, the assembly of a phonetic plan corresponding to the phonological plan, and the implementation of the plan as a speech-motor program (Ozanne, 2005). The creation of a phonological plan necessarily involves a phonological/linguistic component while the remaining levels are motoric in nature. Shriberg and colleagues (2012) view CAS as a multiple domain disorder that includes deficits in *auditory-perceptual encoding*, *memory processes* (i.e., the storage and retrieval of the representations), and *transcoding* (i.e., the transformation of acoustic representations into motor speech movements Shriberg et al. (2017) note that "for speakers of any age with CAS, treatment goals would more appropriately focus on deficits in subdomains of both representational and transcoding processes" (p. S1167). Thus, all three models reviewed above cite deficits in representational aspects of phonemes in CAS that share commonalities with RD. Herein, RD will refer to difficulty acquiring the sound-symbol relationships needed for accurate and fluent word-level reading.

1.2 Models of Word-Level Decoding

Like SSD, RD has theoretical models to explain deficits in reading behavior. Models are based on either a core cognitive deficit such as phonological awareness and lexical retrieval or on multi cognitive issues that combine a cluster of deficits (Fletcher et al., 2019). Regardless of the theoretical model, dyslexia is considered a "*word-level reading disability*" (Fletcher et al., 2019, p. 109). This is consistent with Gough and Tunmer's (1986) "*simple view of reading*," which proposes that difficulties with word-level decoding impede access to meaning— "if print cannot be decoded into language, then it cannot be understood" (p. 7).

Fletcher et al. (2019) provide an overview of multideficit and singular cognitive deficit theories that underlie word-level reading difficulties. For example, the dual-route reading theory proposes subtypes of RD based on reading behaviors that were observed in premorbidly literate adults after suffering a cerebrovascular accident or stroke (Castles and Coltheart, 1993). Phonological dyslexia is proposed to arise from damage to the left perisylvian cortical brain regions, affecting the ability to read words that have consistent grapheme-phoneme correspondences. Surface dyslexia is proposed to occur with anterior temporal lobe damage, characterized by difficulty reading irregular words (e.g., yacht) (Woollams, 2014). Observations of reading behavior in brain-damaged adults led to a *developmental* form of the dual-route theory of RD similarly characterized by phonological and surface dyslexia (Peterson et al., 2013).

Harm and Seidenberg's (1999) connectionist model of developmental dyslexia proposes subtypes of RD that originate from a *single* source—an impairment in the phonological network based on impoverished phonological representations. *Pure phonological dyslexia* is caused by a mild impairment in the phonological system. *Mixed or relative phonological dyslexia* is caused by substantial impairment in the phonological network and affects both irregular and nonword reading (Harm & Seidenberg, 1999; Seidenberg & McClelland, 1989). In the connectionist model, surface dyslexia is regarded as a developmental delay characteristic of children who have had less reading experience.

However, the proposition of pure subtypes of developmental dyslexia has not been well supported. In a study involving 437 dyslexic children, Peterson et al. (2013) evaluated the predictions of the dual-route and connectionist models. While some support was found for

separating groups "at two ends of the distribution," problems surfaced for both the dual-route and connectionist models as the subtypes did not represent independent categories (p. 36). Snowling et al. (1996), reporting on a longitudinal study of 20 dyslexic children and reading-age matched controls, found that poor readers had difficulties reading both nonwords and exception words; furthermore, over the course of the study, the dyslexic children's profile changed, with increasing difficulty reading nonwords, while the controls improved. Snowling (2001) suggested that subtyping of phonological and surface dyslexia may not be a fruitful endeavor because pure subtypes of dyslexia are uncommon, and "taxonomies leave a substantial number of children unclassified" (p. 42).

Research examining how individuals perform on cognitive, linguistic, and perceptual tasks, in addition to poor word-level reading, has also been used to better understand varying subtypes of RD. Morris et al. (1998) analyzed measures of cognitive and linguistic abilities using cluster analysis in a sample of 232 children identified as having a disability in reading, math, reading and math, and three additional groups with diagnoses of ADHD, below-average IQ, or neurotypical controls. Among the reading disabled children, they identified seven subtypes of RD; however, "virtually all children—and subtypes—share[d] impairments on measures of phonological processing" (p. 367). Variability on other components of phonological processing and measures of language and cognitive skills further delineated the subgroups.

1.3 Phonological Processing and RD

The findings of Morris et al. (1998) are consistent with a prevailing theory of RD, the phonological-core-variable-deficit hypothesis, which states that poor readers may have

heterogeneous cognitive profiles, but there is most often a core deficit in phonological processing (Stanovich, 1988). In other words, despite other contributing factors, children with RD appear to suffer from a core cognitive deficit within the phonological component of language (Boada & Pennington, 2006; Cabbage et al., 2018; Goswami, 2000; Peterson & Pennington, 2012; Ramus & Szenkovits, 2008, 2009; Shankweiler et al., 1999; Share & Stanovich, 1995; Snowling, 1995, 1998; Stanovich, 1988; Stanovich et al., 1984; Wagner & Torgesen, 1987). The working definition of dyslexia as formulated by the National Institute of Child Health and Human Development emphasizes this point:

> Dyslexia is a specific learning disability that is neurobiological in origin. It is characterized by difficulties with accurate and/or fluent word recognition and by poor spelling and decoding abilities. These difficulties typically result from a deficit in the *phonological component of language* that is often unexpected in relation to other cognitive abilities and the provision of effective classroom instruction. (Lyon et al., 2003, p. 2, emphasis author's)

While a deficit in the phonological component of language may surface as difficulty with word-level decoding (mapping speech sounds onto letters), it can be measured behaviorally by phonological processing skills, such as the following:

- **phonological awareness (PA)**: a meta-linguistic skill demonstrated by the ability to synthesize and manipulate sounds in words (Bradley & Bryant, 1983; Fletcher et al., 1994; Torgesen, & Burgess, 1998; Wagner et al., 1997)

- **phonological memory (PM)**: the ability to encode and repeat verbal material of increasing length (Mann & Liberman, 1984; Torgesen, 1985)
- **lexical retrieval (RAN)**[1]: the ability to quickly retrieve phonological information from long-term memory (e.g., rapid automatic naming of objects, colors, digits, or letters) (Bowers, 1995; Christo & Davis, 2008; Norton & Wolf, 2012)

Phonological processing skills are proposed to rely on intact phonological representations, and a widely accepted hypothesis of phonological processing difficulties is that degraded or underspecified phonological representations undermine efficient processing (Ramus & Szenkovits, 2008). However, Bishop and Snowling (2004) emphasize the importance of "unpacking" concepts of phonological processing to disentangle the component skills contributing to the construct (p. 879). For example, when considering PA tasks such as phoneme manipulation, difficulties may surface because of differing issues: underspecified phonological representations, rapid decay of representations, access, executive function, and memory skills. Further complicating our understanding of phonological processing, Ramus and Szenkovits (2008) argue that difficulty in phonological processing tasks, such as multisyllable word repetition or phoneme manipulation tasks, lies in *access* to the target phonological information rather than degraded representations. Results from a series of experiments involving phonological processing tasks for college-age students with RD indicated that phonological

[1] Herein, PA will be used to refer to phonological awareness, PM will refer to phonological memory, and RAN will refer to rapid automatized naming unless the terms apply to names in a published test or appear in cited material.

processing difficulties surfaced only in tasks that relied heavily on short-term memory, conscious awareness, and time constraints (Ramus & Szenkovits, 2008).

Finally, differences in deficits in the components of phonological processing have been proposed to describe subtypes of RD. For example, Lovett (1987) suggested that young readers with accuracy-related deficits exhibited oral language deficits and difficulty with "speech sound analysis," while children with rate-related deficits (i.e., slow but accurate readers) had specific deficits in naming speed. Wolf and Bowers (1999) extended Lovett's profiles of rate and accuracy to advance the double deficit hypothesis. They proposed that children with word-level decoding deficits can have single deficits in either speech sound analysis (i.e., PA) or rate (i.e., processing speed, RAN) *or* simultaneous difficulty with both PA and RAN. Individuals with a double deficit are proposed to have more severe reading difficulties.

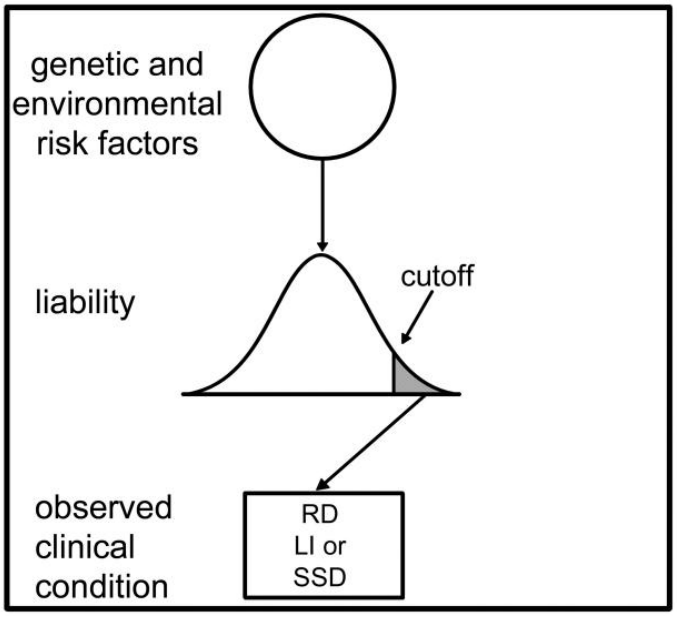
1.3.1 The Association of Phonological Processing, RD, SSD, and LI

Phonological processing deficits are a feature in all the theoretical models of SSD and RD discussed above. Given this commonality, it is not surprising that SSD and RD are often comorbid, with overlap at behavioral, neural, and genetic levels (Bishop & Snowling, 2004; Pennington, 2006; Pennington & Bishop, 2009; Peterson et al., 2007). As such, they are hypothesized to share common risk factors. One way to conceptualize the endophenotypic overlap of these disorders in the context of separate phenotypes is to adopt a model that emphasizes "multiple overlapping risk factors" (Pennington & Bishop, 2009, p. 301). In this conceptualization, independent genetic and environmental factors, both risk and protective, will determine a specific underlying deficit; the observed disorder depends on the combination of

these liabilities that exceeds a given threshold. Figure 1 shows a schematic illustration of a continuous liability distribution; once a certain threshold is met, RD, LI or SSD is observed. For example, one aspect of phonological processing difficulties, how phonemes are represented and stored for use in a child's developing language system, is considered a cognitive component in both SSD and RD. In contrast, deficits in lexical retrieval and access (RAN) are posited to be particular to RD (Pennington & Bishop, 2009).

Figure 1

Continuous liability distribution



Note. RD, reading disability; LI, language impairment; SSD, speech sound disorder. From "Relations among speech, language, and reading disorders," by B.F. Pennington and D. V. M. Bishop, 2009, Annual Review of Psychology, 60(1), 283-306. Copyright (2009) by Annual Reviews Inc. Reprinted with permission.

Consistent with a "multiple overlapping risk factors" model, the manifestation of speech difficulties associated with phonological processing deficits for both SSD and RD can be viewed on a continuum, ranging from subtler difficulties in speech tasks such as multisyllable word

production to overt disorders in articulation (Cabbage et al., 2018; Catts, 1986). For example, difficulties with multisyllable word repetition might be the only speech manifestation of phonological processing deficits for individuals with RD. Repeating multisyllable words requires an individual to encode and then verbally sequence phonologically complex information to reproduce a word. As noted, individuals with RD are proposed to have deficient phonological representations, making storage and retrieval of multisyllable words challenging. Alternatively, as Ramus and Szenkovits (2008) have suggested, difficulties may surface due to accessing the phonological information in a timely way.

Catts (1986) investigated the association of reading ability and multisyllabic word repetition for adolescents with a diagnosis of RD, comparing them to age-matched controls with no reading difficulties. The participants with RD made significantly more errors than the controls repeating multisyllabic words, and performance on the repetition task correlated with reading ability, especially nonsense word decoding. Production errors comprised omissions or substituted sound segments (e.g., ther<u>m</u>ometer > ther ometer, Christmas or<u>n</u>ament > or<u>d</u>ament), *not systematic articulation errors*. Catts concluded that difficulty "forming phonological memory codes, as well as the reactivation/execution of these codes/programs, may underlie RD children's speech production deficits" (Catts, 1986, p. 507). The RD participants in the Catts study are presumed to be similar to the participants in the current study with RD-no SSD, (i.e., presumed to have phonological processing deficits that did not result in an SSD). However, as there was no specific mention of excluding RD participants with a history of SSD, this may not have been the case.

On the other end of a continuum of speech production difficulties associated with deficits in phonological processing, are SSD. One type of SSD described earlier, a phonological disorder, is thought to be the results of cognitive-linguistic issues, i.e., difficulty establishing accurate representations of phonemes used for speech production (Anthony et al., 2011; Fey, 1992; Rvachew & Grawberg, 2006; Rvachew & Jamieson, 1995). Longitudinal studies examining the outcomes for preschool children with a phonological disorder confirm an association of early SSD and delayed acquisition of reading in the elementary grades (Bird et al., 1995; Larrivee & Catts, 1999; Lewis et al., 2004; Nathan et al., 2004; Rvachew, 2007). Estimates of roughly 25% of children with RD demonstrate a history of "clinically significant speech production" issues in preschool (Pennington & Lefly, 2001, p. 830), and 30% of children with a history of SSD obtain a diagnosis of RD at school-age (Lewis, 1996). Raitano et al. (2004) examined the persistence of speech errors in 5- to 6-year-old children with and without comorbid language impairment (LI) and found that the persistence of a speech disorder made an independent contribution to the performance on PA tasks. Even children whose speech had normalized without LI were found to have deficits on PA tasks relative to the control participants.

Rvachew and Grawberg (2006) investigated the association of articulation, PA, speech perception, and emergent literacy in a group of 95, 4- to 5-year-old preschool-age children with SSD. The study demonstrated that children with SSD are at significant risk for delays in developing PA skills and that emergent literacy skills were almost entirely predicted by PA (Rvachew & Grawberg, 2006). In another study, Rvachew (2007) examined the association of PA skills measured before kindergarten entrance and reading skills at the end of first grade in a

group of children with SSD. The children who performed most poorly on PA tests (rime and initial sound recognition) in kindergarten had significantly lower nonword decoding skills than the SSD group and controls, who demonstrated more typical PA skills. In this study, reading ability was correlated with PA skills and not SSD.

Adolescents with persistent SSD have also been found to have weaknesses in phonological processing and comorbid issues with reading and spelling. Preston and Edwards (2007) used multisyllable word repetition tasks and phoneme manipulation tasks to compare the phonological processing skills in older school-age children and adolescents with residual speech sound errors to those adolescents of the same age and language abilities with no speech sound errors. The group of participants with SSD performed significantly more poorly on five of the six phonological processing tasks. The majority of the participants in the SSD group received intervention for reading and spelling.

Lewis et al. (2015) investigated the outcomes for adolescents with an early history of SSD. The study included three groups: a resolved-SSD group with normal articulation in conversation and average performance in multisyllable word repetition (MSW), a persistent-SSD group with inaccurate speech sound production in conversation and difficulty with MSW, and a low-MSW who continued to demonstrate difficulty with multisyllable word repetition but had no overt speech errors in conversation. While language and literacy scores were lowest for the Persistent-SSD group, deficits in phonological processing alone (low-MSW group) predicted lower scores across all language and literacy measures. Thus, an early SSD, even for adolescents

whose SSD has resolved (i.e., no overt speech sound errors), poses a significant risk for language and literacy problems.

There is also evidence that children with RD, in addition to a core deficit in phonological processing and possible difficulties with SSD, suffer from broad-based linguistic deficits—LI (Bishop & Adams, 1990; Kamhi & Catts, 2012; Lombardino et al., 1997). McArthur et al. (2000) investigated the overlap between RD and LI in 212 children who had received a diagnosis of either RD or LI. Fifty-five percent (61/110) of the children diagnosed as RD also demonstrated LI. Fifty-one percent (52/102) of the participants with LI were also diagnosed with RD. The high comorbidity rate raises questions about a common underlying deficit in these two disorders.

Plaza et al. (2002) proposed the *processing limitation hypothesis, as* tying both LI and RD together. Inefficient processing of phonological structures cause limitations in phonological memory and are hypothesized to affect both spoken and written language. The 26 study participants with RD demonstrated significant concurrent impairment in oral language (i.e., word retrieval, verbal short-term memory, syntactic processing, and semantic production) compared to both age-matched and younger-age controls. The participants demonstrated the greatest difficulty in repeating unfamiliar multisyllabic words due to difficulties with encoding and retrieving the articulatory movements corresponding to the encoded phonological information.

Catts et al. (1999) investigated the contribution of oral language and phonological processing skills to RD in a longitudinal study of 183 kindergarten children later diagnosed with RD. They found that kindergarteners' oral language and phonological processing skills predicted

reading in second grade. Furthermore, oral language and phonological processing made unique contributions to single-word decoding. In two follow-up studies, Catts et al. (2005) investigated the overlap between LI identified in kindergarten and dyslexia identified in 2nd, 4th, or 8th grades and the contributions of phonological processing to LI and RD. According to their findings, there was limited but significant overlap between LI and RD. Specific phonological processing and decoding issues characterized RD, whereas LI involved semantics, syntax, and language processing deficits. While the two disorders are often comorbid, a problem in phonological processing does not appear to be a significant factor in LI when it occurs in isolation. Their results support a view of LI in which a deficit in phonological processing is closely associated with LI only if it occurs with a comorbid diagnosis of dyslexia.

1.3.2 The Association of Phonological Processing, RD, CAS, and LI

CAS is a subtype of SSD whose cause is thought to originate in difficulty accessing and executing motor plans for speech production (ASHA, 2007). As described earlier, CAS can be distinguished from a phonological disorder by the putative underlying cause, i.e., motor-speech planning versus cognitive-linguistic (phonological) deficits, and, in the often, persistent nature of the speech disorder. The following is ASHA's official policy statement for CAS:

> Childhood apraxia of speech (CAS) is a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone). CAS may occur as a result of known neurological impairment, in association with complex neurobehavioral disorders of known or unknown origin, or as an idiopathic

> neurogenic speech sound disorder. The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody (ASHA, 2007, pp. 3–4).

ASHA's characterization of CAS as primarily affecting "speech sound production and prosody" belies the fact that children with CAS often present with a broad range of comorbid deficits, including literacy problems (Aram & Nation, 1982; Gillon & Moriarty, 2007; Lewis et al., 2004; Marion et al., 1993; Marquardt et al., 2002; McNeill et al., 2009a; Miller et al., 2019; Stein et al., 2020; Zaretsky et al., 2010). While there is abundant research about the comorbidity of a phonological disorder, phonological processing deficits, and RD, there is a limited amount of research about these same issues in CAS (Bird et al., 1995; Bishop & Adams, 1990; Preston et al., 2013; Raitano et al., 2004).

Moreover, the "critical age hypothesis", which proposes that persisting speech difficulties at the time children are first learning to read (i.e., 5 to 6 years of age) undermines subsequent literacy skills (Bird et al., 1995; Bishop & Adams, 1990; Hayiou-Thomas et al., 2017) seems particularly applicable for children with CAS. In an extension of the critical age hypothesis, Nathan et al. (2004) suggested that poorly specified phonological representations underlie both persistent speech errors and phonemic awareness. Children with more severe and persistent speech problems, such as in CAS, are therefore more likely to have difficulty learning to read (Bird et al., 1995; Nathan et al., 2004).

Marion et al. (1993) proposed that an underlying phonological processing deficit may be causative in CAS. The investigators found significant deficits in rhyme generation and rhyme identification in their study of 5- to 7-year-old children with developmental apraxia of speech (DAS). According to their conceptualization, motor-speech difficulties were attributable to ill-formed phonological representations required to guide articulatory output. They concluded that both CAS and developmental dyslexia share a neurological basis in which affected children may not be able to master the conversion of graphemes to phoneme associated with fluent reading. Marquardt et al. (2002) also viewed CAS as involving an impoverished phonological representation system, finding deficits in phonological processing for all three participants with CAS in their study of metalinguistic tasks of syllable perception. The authors concluded that DAS is not a simple motor control deficit but has linguistic underpinnings.

Several longitudinal studies have charted the developmental trajectory of phonological processing, oral language abilities, and literacy for school-age children and adolescents with CAS (Lewis et al., 2004; Stackhouse & Snowling, 1992). Stackhouse and Snowling (1992) evaluated the persistence of speech errors, phonological processing abilities, and literacy outcomes in a longitudinal study that followed two school-age children with CAS into adolescence. In addition to significant difficulties with speech production, both participants demonstrated problems with rhyme production and rhyme recognition, sound blending, and sound discrimination tasks at 14 to 15 years of age. The findings suggest that persisting speech difficulties for children with CAS arise from a complex interaction of phonological representations, lexical representations, and motor planning difficulties. The authors proposed

the term “developmental verbal dyspraxia” (DVD) rather than developmental articulatory dyspraxia or dyspraxia of speech because of the combination of “pervasive spoken and written language problems” (p. 53).

Studies also confirm increased rates of LI relative to population estimates and other forms of SSD for children with CAS, although many of these studies focused primarily on expressive language deficits (Ekelman & Aram, 1983; Thoonen et al., 1997). Thoonen et al. (1997) reported that 82% of their CAS participants had a comorbid language impairment; however, no differentiation was made between receptive and expressive language in the study. If motor-speech impairment is the central deficit of CAS, then expressive language skills should improve as speech intelligibility improves (Hall, 1992). This expectation was not borne out in Lewis et al. (2004), who compared the outcomes at school age for children diagnosed with CAS to those with either an isolated SSD (S group) or a combination of SSD and language disorder (SL group) in early childhood. While speech difficulties in the CAS group decreased over time, expressive language difficulties persisted, with 90% of their school-age participants with CAS demonstrating expressive language deficits. These findings are consistent with Murray et al. (2019), who found that preschool children with a CAS diagnosis had difficulties with expressive grammar unrelated to speech production difficulties.

Additionally, the superiority of receptive language skills over expressive language is considered a hallmark of CAS (Rosenbek & Wertz, 1972; Velleman, 2003). Thus, despite speech production difficulties, receptive language should remain relatively intact. In one of the few studies that examined receptive language, Lewis et al. (2004) reported that most of their sample

of school-age participants with CAS demonstrated comorbid receptive language deficits. Seventy percent of the children with CAS scored 1 *SD* or more below the mean (85) on the receptive language portion of the Clinical Evaluation of Language Fundamentals-Revised (CELF-R). The CAS group was also more impaired than the other two groups in reading. Spelling was particularly impaired in the CAS group, signaling difficulty in speech sound analysis and segmentation abilities, perhaps due to constraints in PM (Snowling & Stackhouse, 1983).

Several case studies shed light on the developmental trajectory and association of speech sound production, phonological processing skills, and decoding for children with an early diagnosis of CAS. In a longitudinal study of a clinical case, Zaretsky et al. (2010) described the speech, language, and literacy outcomes for a school-age participant with severe CAS and borderline IQ from 6 through 11 years of age. Deficits in speech production, expressive and receptive language, and phonological processing skills persisted despite intensive intervention. Furthermore, the study proposed that deficits in PA, PM, and working memory for children with CAS underlie difficulties with decoding.

Another single-case clinical study examined the outcomes in speech production, oral language, phonological processing, and literacy abilities for a 15-year-old adolescent male diagnosed with CAS (Turner et al., 2019). Speech, language, literacy, and academic outcome data were collected from ages 3;10 to 15 years. At school age, PA skills were measured to be in the average range; however, PM abilities fell 2 *SD*s below the mean. By 15 years of age, the subject had acquired intelligible speech and average-range receptive and expressive language skills, but deficits in reading comprehension, decoding of nonsense words, and spelling persisted.

Preston et al. (2016) investigated persisting speech errors for individuals with CAS in a treatment study involving biofeedback. Three participants diagnosed with CAS ranging in age from 10 to 13 years underwent treatment for the persistent distortion of the /ɹ/ sound. All three participants demonstrated significant deficits in PA and oral language. While improvement was noted for treated sounds, none of the three participants made progress in generalizing the correct production of their target sounds to untrained contexts during the intervention. The authors concluded that phonological processing deficits may have impeded progress due to a lack of "training on the proper acoustic target," underscoring the importance of accurate phonological representations for sound production (p. 378).

Miller et al. (2019) investigated word-level decoding skills and speech-language correlates for school-age children and adolescents between 7 and 18 years of age with an early diagnosis of CAS (N = 40), or a diagnosis of speech sound disorder but not CAS (SSD-no CAS; *n* = 119). Of the CAS participants, 65% qualified as below-average decoders compared to 24% of the SSD-no CAS. As predicted, oral language abilities and phonological processing skills were related to literacy skills, similar to children with other forms of SSD. In another retrospective study, Miller et al. (in preparation) investigated speech sound production, decoding, and PA skills for a group of participants (N = 32) diagnosed with CAS, ages 12 through 25 years. Fifty-nine percent (19/32) of the participants demonstrated persistent speech sound errors beyond 12;6 years. Regardless of speech persistence, participants demonstrated weaknesses in decoding, PA, and complex speech production tasks. Over half of the participants scored below one standard deviation of the mean on a word-level decoding assessment at the final evaluation.

Furthermore, the means for both groups of participants, persistent and not persistent, fell below the normative average on a measure of PA.

1.4 Models of Speech Perception and Speech Production

Speech perception is the process by which acoustic information is transformed into linguistic representations (Rvachew & Grawberg, 2006). The robustness of this process is essential to the development of accurate phonological representations and lays the foundation for spoken and written language (Breier et al., 2002; Goswami, 2014; Hearnshaw et al., 2019; Molfese, 2000). For instance, a 6-month-old infant's ability to discriminate between two vowels significantly predicts language outcomes at 2 years of age (Tsao et al., 2004). Furthermore, an infant's vowel perception at approximately 8 months predicts pre-literacy skills and language abilities at the age of 5 (Cardillo-Lebedeva & Kuhl, 2009). However, there are differing views of how acoustic information is transformed into phonetic and then phonemic categories (Rvachew & Grawberg, 2006). Hickok et al. (2011) describe two differing accounts of the process: the *motor-centric view* and the *audio-centric view* (p. 407).

The motor theory of speech perception (1985), a motor-centric view proposed by Liberman and Mattingly, asserts that individuals perceive speech by a human-specific, innate module that recovers the intended vocal tract gesture (phonemes) from the acoustic output. The Direct Realism Theory (Fowler, 1986), also a motor-centric view, asserts that vocal tract gestures form the basis for speech perception, but unlike Liberman and Mattingly's version, there is no innate module. Listeners directly "perceive" vocal tract gestures from acoustic information as these are common between the listener and the speaker. The motor theories purport to solve the

problem of "contextual dependence"—that an auditory signal associated with speech sounds can be variable, but the motor gestures that produce them are not (Hickok et al., 2011).

In accordance with the motor theory, children with SSD might have difficulty creating correct articulatory movements because of their inability to process or recover the gestures associated with the phonemes (Namasivayam et al., 2020). However, Imada et al. (2006) demonstrated that the link between phonetic perception and activation in the infant speech motor cortex develops over time, which is not consistent with the idea that speech perception involves the recovery of innate articulatory gestures. Additionally, the ability to perceive speech sounds has been demonstrated in individuals who were never able to speak due to a congenital disease.

Bishop et al. (1990) compared children and adolescents between the ages of 10 and 18 years of age with cerebral palsy (i.e., anarthria or severe dysarthria) to age- and nonverbal ability-matched controls. The individuals with speech impairment did less well than controls on a phoneme discrimination task of nonwords and receptive vocabulary. However, when memory constraints were removed by adding pictures and making it a word judgment task, the individuals with speech impairment performed as well as controls with phoneme contrasts. Individuals with speech impairment did not appear to have a reduced set of phoneme contrasts; rather, they appeared to have difficulty differentiating nonwords, which may require overt and covert rehearsal.

The audio-centric view of speech perception maintains that the auditory system assumes the central role in perception (and production) (Hickok, 2011). Contrary to the assertions of the

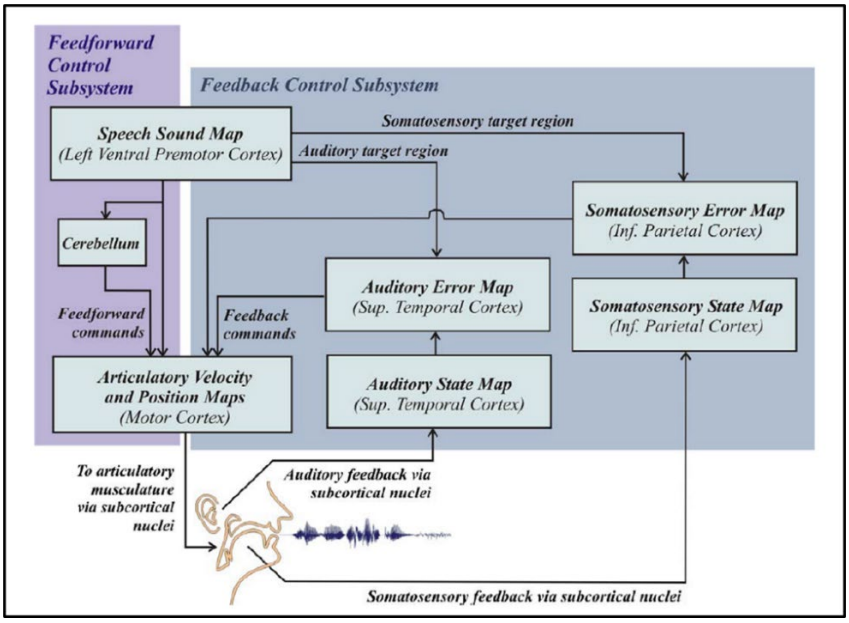
motor-centric view of speech perception, there is a lack of invariance in the articulatory positions used to produce speech sounds; thus, an auditory target has been proposed as the basis of speech perception (Kearney & Guenther, 2019). Neurocomputational models of speech processing attempt to reconcile the auditory and motor theories of speech perception but are largely audio-centric developmentally. For example, the mapping from Directions in sensory space Into Velocities of Articulators (i.e., DIVA) model integrates auditory (acoustic signal) and somatosensory (i.e., tactile and proprioceptive) information from the vocal tract, with internal models into a cohesive account of adult speech production (Guenther, 2016; Guenther & Vladusich, 2012; Kearney & Guenther, 2019). It is consistent with theories of speech production in which auditory targets guide phonetic planning and articulation. The model strives to be neuroanatomically plausible, with each process of the model posited to reside within a neuroanatomical area of the brain and generating analyzable articulatory and acoustic data via a speech synthesizer to make predictions (Terband, 2014) .

The DIVA model begins with the speech sound map, a neuroanatomical area proposed to be responsible for the basic motor plans for speech production. In Figure 2. The speech sound map corresponds to the *mental syllabary,* a component of Levelt's *lexical access model* (Figure 3). The *lexical access model* is an earlier model of speech production that describes stages of lexical access not represented in the DIVA model (Kearney & Guenther, 2019; Levelt, 1992; Levelt, 1994). The model involves conceptual preparation (forming an idea), lexical access (finding the word that represents the idea), grammatic encoding (incorporating grammatic elements), phonological word encoding (sequencing of phonological information), phonetic

planning (associating the motor plan with the phonological sequence) and finally the articulation of a target message. The phonological word encoding stage assembles a sequence of phonological elements. The mental syllabary translates "an abstract phonological representation of an utterance into a context-dependent phonetic representation which is detailed enough to guide articulation" (Levelt, 1994, p. 240). Difficulties with phonetic planning and programming, observed in the adult form of apraxia of speech (AOS), correspond to the mental syllabary and the DIVA model's speech sound map, hypothesized to be located in the left ventral premotor cortex (Guenther, 2016).

Figure 2

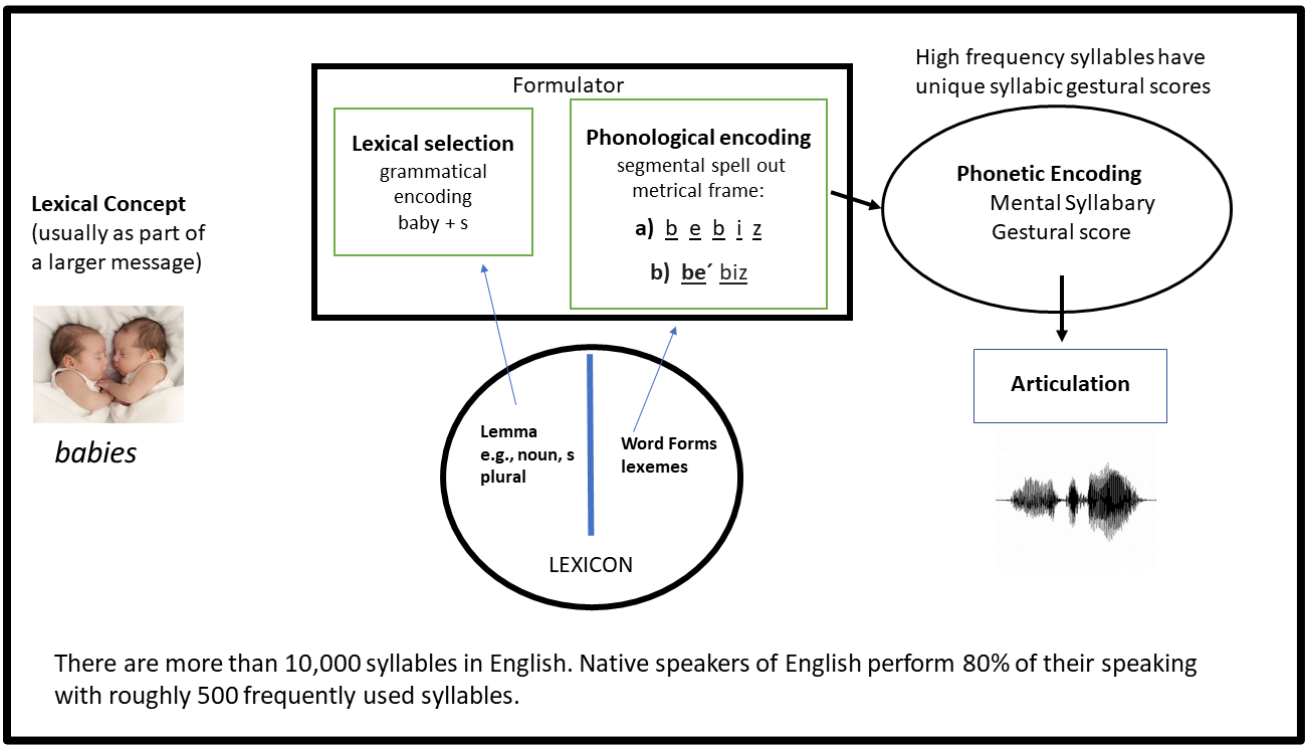
Hypothesized neural processing stages involved in speech acquisition and production according to the DIVA model



Note. Projections to and from the cerebellum are simplified for clarity. Reprinted from "Neural modeling and imaging of the cortical interactions underlying syllable production," by F. H. Guenther, S. S. Ghosh, J. A. Tourville, 2006, *Brain and Language*, p. 282. Copyright 2005 by Elsevier Science & Technology Journals. Reprinted with permission.

Figure 3

An outline of lexical access in speech production



Note. Adapted from Accessing words in speech production: Stages, processes, and representations by W. J. M. Levelt, 1992, *Cognition*, 42, p. 4. Copyright 1992 by Elsevier Science B.V. Adapted with permission.

The current DIVA model provides some limited explanation for the developmental phases of speech acquisition and production. The following is a summary of the basic components of the developmental phases of the model (Guenther, 2006; Terband et al., 2014). During the first phase of speech development (i.e., the systemic mapping stage), semi-random movements of the articulators are paired with auditory and somatosensory feedback. In this stage, an infant learns the relationships between somatosensory and auditory feedback to create systemic mappings and motor commands. During the second phase, the babbling stage, the child

learns more reliably what movements are required to produce an auditory target and what auditory outcome will occur with specific motor configurations. During the next phase, the imitation stage, also known as the phoneme/phonological phase, the child imitates target sounds relying heavily on the auditory feedback control subsystem. The feedforward command incorporates the error corrections based on the auditory feedback control subsystem for each attempt. It then updates the feedforward control, resulting in a more accurate feedforward command on the next attempt (Guenther, 2006). The final phase is mature speech production, in which the feedforward commands are accurate enough for the model to produce target sounds without generating auditory errors (Terband et al., 2014). The DIVA model predicts a causal relationship between the acquisition of the auditory coordinates of speech sounds and their associated motor programs. Guenther and Vladusich (2012) propose that "individuals with more distinctive auditory speech representations—those people better able to discriminate between similar speech sounds—should produce more distinctive speech utterances than those with poorer auditory discrimination (p. 10).

1.4.1 Speech Perception and SSD

Research has been conducted specifically about the influence of speech perception on production in SSD. Shiller et al. (2010) reviewed 14 papers published in ASHA journals since 1951 on the association of speech perception and phonological disorders. The authors reported that 13 of the 14 studies demonstrated "unequivocal evidence for significantly poorer speech perception abilities on the part of children with delayed speech" (p. 184). Shiller et al. posited that this association relates to difficulties with auditory to phoneme mapping, creating poor auditory models leading to imprecise and inaccurate articulation.

A more extensive systematic review, which included 73 studies, and a meta-analysis (a subset of eight studies) dating from 1931 through 2016, investigated the association of speech perception and SSD in preschool and early school-age children (Hearnshaw et al., 2019). Participants comprised children with SSD (including those with CAS) who ranged in age from 3;0 to 8;2 years. Out of 73 studies, 60 found that some or all the SSD participants had difficulties with speech perception compared to controls. The meta-analysis revealed that children with SSDs performed more poorly on lexical and phonetic judgment tasks than children with typically developing speech in the subset of all eight studies. Unfortunately, information about speech perception skills based on SSD subtypes was not possible due to terminological changes across the 85 years of the reviewed literature (Hearnshaw et al., 2019). This lack of clarity has implications for the conceptualization of specific diagnostic categories in SSD. For example, some children diagnosed with motor-speech disorders (i.e., CAS) may have had difficulty with speech perception, implicating higher-level linguistic deficits, similar to other subjects with SSD.

There are also differing theoretical positions about the direction of influence of speech perception and production in SSD. Most of the evidence reviewed by Hearnshaw et al. (2019) supports speech perception deficits lead to inaccurate speech production, consistent with Shiller et al. (2010). However, several other positions have also been offered. A position partially compatible with Liberman and Mattingly's motor theory proposes that speech production influences speech perception, primarily for a child's error sounds. For example, Ohde and Sharf (1988) found that the "perceptual problem of children with an articulation error relates to the abnormal production of this sound and not to a global deficit in perception" (p. 567). Wolfe et al.

(2003) found that production training for error sounds alone significantly influenced these sounds' identification (perception). However, production training alone was not as successful as mixed training (production and identification) for error sounds that children had little perceptual knowledge of at the beginning of the intervention.

A bi-directional influence of speech perception and production has also been suggested (Borrie & Schafer, 2015; Byun, 2012; Rescorla & Ratner, 1996). For example, in the DIVA model, the auditory target is the primary target for speech production early in development (Guenther, 2016); however, the auditory target is supplemented by somatosensory information, and eventually, both auditory and somatosensory information underpins the mappings for speech sounds. Kuhl and Meltzoff (1982) propose that infant speech perception is an intermodal event comprising the "intermodal organization of auditory, visual and motor concomitants of speech," the intertwining of which are especially "conducive" to vocal learning and production (p. 1140). Children with SSD are hypothesized to have an "underlying phonemic inadequacy [and] may elect to vocalize less" (Rescorla & Ratner, p. 163, 1996), thus reducing opportunities for vocal practice thought to promote phonological development (Stoel-Gammon, 2011).

Speech perception is also reported to influence PA skills for children with SSD (Benway et al., 2021; Rvachew et al., 2003; Rvachew & Grawberg, 2006). Rvachew et al. (2003) examined PA and speech perception skills in two groups of 4-year-old children, one with typical speech and language skills and one with moderate to severe phonological delays. The children with expressive phonological delays demonstrated significantly poorer speech perception and PA skills than the controls. Benway et al. (2021) examined speech perception in a group of 110

school-age children with persistent /ɹ/ distortions using an identification task of the child's error sound. They also assessed receptive vocabulary and PA skills. Findings supported the direct relationship between auditory perceptual acuity and PA for these school-age children with residual speech sound errors. Children who were more consistent in assigning tokens between the /ɹ/ and /w/ categories to the correct phoneme category on a synthetic rake–wake continuum had better PA composite scores. Tsao et al. (2004) demonstrated that speech perception measured at 6 months of age by the ability to discriminate between two vowels in a head turning task, was the most important predictor of lexical production at 24 months.

1.4.2 Speech Perception and CAS

The bidirectional influence of speech perception and production described above is particularly relevant for children with motor speech production difficulties. Infants with motor-speech disorders, such as CAS, demonstrate decreased speech-like vocalization during infancy (Overby, 2015; Velleman, 2011). Highman et al. (2008) investigated parents' recollections of their children's babbling for typically developing infants (TD), infants with suspected CAS, and infants with specific language impairment (SLI), i.e., language difficulty cannot be accounted for by general delay in development, hearing loss, oral structural issues, ASD, apraxia, or acquired brain injury. Thirty-five percent of the infants with suspected CAS were reported to demonstrate no babbling, whereas 100% of the infants in the SLI and TD groups were reported to babble. Additionally, 95% of the infants later diagnosed with CAS exhibited an absence of variegated babbling compared to infants in the SLI and TD groups. The limitation of verbal output and attenuation of more mature babble suggests underlying bidirectional difficulties with speech perception and production. The overall effect of which reduces opportunities for practice

forming auditory, phonological, and somatosensory connections (representations) required for accurate speech sound production (Velleman, 2011).

To further investigate the interaction of speech production and perception during early speech development for children with CAS, Terband et al. (2014) used the DIVA computer model to simulate auditory and motor processing deficits during the babbling and imitation stages of speech development. Their findings revealed a close relationship between poor auditory self-monitoring (auditory processing) and motor deficits (APD+MPD), leading to simulated verbal output, characteristic of CAS symptoms. Motor deficits alone (MPD) produced an SSD with only a phonological component. Terband et al. (2014) concluded that "the auditory perception impairments that have been found in children with CAS are not derived or consequential, but that the deficit in auditory processing might play a fundamental role in the emergence of the disorder" (p. 29).

Acoustic studies have also investigated speech perception deficits as a potential cause for speech difficulties and related language and reading difficulties for children with CAS. Groenen et al. (1996) assessed 17 children with CAS (*M* = 8;9 years) and 16 controls (*M* = 8;0 years) using a discrimination task (auditory memory and phonetic information) and an identification task (phonemic judgment based on phonetic information). The stimuli consisted of resynthesized and synthesized monosyllabic words differing in place-of-articulation and alterations of the intensity of the third formant of the initial voiced stop consonant (e.g., dak versus bak - real words in Dutch). The identification task demonstrated equal slopes for both groups, indicating no deficit in phonetic processing for the children with CAS. However, the CAS participants showed

poorer discrimination of same/different judgments than the controls. Additionally, the discrimination performance and the speech errors of the CAS participants were related; CAS participants demonstrated a specific relation between discrimination ability and the number of place-of-articulation substitution errors. Groenen et al. (1996) concluded that "the presumed neurological defect that initially disrupted the oral-motor functioning of the child may also have disrupted the auditory functioning of the child at the same time" (p. 479).

Maassen et al. (2003) investigated the auditory and phonetic processing abilities of 11 children with CAS and 12 controls with normal hearing, cognitive, motor and perceptual functioning, without speech and language problems. They reduced the formant frequencies for two vowel continua—/i/ (P<u>ete</u>) versus /I/ (p<u>i</u>t), and /ɑ/ (<u>o</u>n) versus /ʌ/ (b<u>u</u>n), which were then used in an identification task. An AX same/different discrimination task was also used with the same vowels. Results indicated poorer perception and discrimination of vowels for the children with CAS. The authors concluded that the internal representations of vowels seemed poor, which affected both production and perception.

In another study of speech perception, Nijland (2009) examined auditory perceptual abilities in children with CAS, phonological disorders (PD), mixed disorders (characteristics of both CAS and PD), and controls. Children with CAS did not differ from children with PD on the higher-order perceptual skills of rhyming and categorization; both did poorly compared to the control group. However, children with CAS demonstrated deficits in the lower-order perceptual skills task of non-word discrimination, unlike children with PD, who scored similarly to controls. Additionally, perception (non-word discrimination and rhyming) and production of words and

non-words were correlated for the children in all three groups with articulation issues; the better the speech productions scores were, the higher the discrimination and rhyming scores were. Nijland and colleagues concluded that children with CAS show auditory perceptual deficits for higher- and lower-order linguistic input; difficulty with lower-order processes like speech discrimination may create a bottom-up effect on higher-order phonemic mapping. Bishop's (1997) characterization of developmental disorders as "a complex pattern of associated impairments . . . an impairment at an early stage of processing affects all the processes downstream of that stage" seems consistent with the hypothesized effects of lower-level perceptual deficits affecting higher-level phonemic processing found in this study (p. 904).

Zuk et al. (2018) investigated deficits in speech perception for children with CAS with and without accompanying LI. Comparison groups included children with speech delay only, LI only, and typically developing children. Children with CAS+LI showed significantly poorer syllable discrimination abilities (i.e., /dɑ/–/gɑ/ syllable contrast in the third formant frequency associated with the consonant's place of articulation) compared to children with CAS-only and typically developing peers. However, children with speech delay also showed poorer discrimination abilities but had substantial within-group variability. The authors concluded that speech perception deficits are not a primary feature of CAS but occur with comorbid LI. There were several limitations of the study, including the broad age range of participants (i.e., from 4 through 17 years). Additionally, in some cases, the group with speech delay demonstrated poorer speech perception than both CAS groups, signaling the possibility that speech perception issues are associated with speech production difficulties regardless of SSD category or language status.

Vowel duration is a critical feature of lexical stress (Peter & Stoel-Gammon, 2005). Vowel errors and prosodic disturbances (i.e., equalized stress and syllable segregation) are hallmarks of speech production difficulties for children with CAS. To better understand these prosodic disturbances and vowel errors, Ingram et al. (2020) evaluated the ability of nine children with CAS and 14 typical controls, ages of 5 through 6;11 years, to discriminate pairs of the /bɑ/ syllable differing only by vowel duration. Overall, children with CAS were less accurate in determining vowel duration, demonstrated by greater variability across duration difference conditions, and a flatter slope with smaller increases in discrimination improvement as the durations increased.

In one of the few speech perception experiments using competing masking noise, Iuzzini-Seigel et al. (2015) investigated the stability of feedforward commands in school-age children with CAS, SD (speech delay), and TD controls between the ages of 6;1, and 17;6 years. Speech development in its earliest stages is hypothesized to rely on auditory and somatosensory feedback to create internally represented, stable motor plans for speech output; these are known as feedforward commands consistent with the DIVA model (Guenther, 2016). Feedback commands become increasingly less important as a young child fine-tunes motor plans; with development, feedback commands are used only when speech output becomes perturbed in some way. It is hypothesized that robust feedforward programs, assumed for children with typical speech development, result in accurate output even when noise disrupts access to auditory feedback—an overreliance on feedback signals weakly established feedforward motor commands.

Participants in the Iuzzini-Seigel study repeated consonant-vowel-consonant (CVC) pseudowords, /pɑb/, /pæb/, /pib/, and /pub/ presented with and without masking (white noise presented at 65 dB). The participant's response was produced concurrently with the masking noise, thus eliminating any possible auditory feedback. Speech output was analyzed for VOT (voice onset time), percent VOT correct, vowel length, vowel space, and speech intensity. Children with CAS produced fewer correct VOTs for targets and reduced vowel space area when auditory feedback was masked; in contrast, the SD and TD groups showed no masking effect on any measures. The authors concluded that the participants with CAS had not developed robust feedforward programs and were disadvantaged by the masking noise, unlike the SD and TD groups.

Froud and Khamis-Dakwar (2012) used neurophysiological methods to investigate the hypothesis that children with CAS demonstrate phonological over-specification. The authors used high-density EEG (electroencephalography) to measure responses during a Mismatched Negativity Response (MMN) task. MMN responses were elicited during two passive oddball paradigms in which standard repetitive stimuli were presented interspersed with deviant stimuli (standard - /pa/ vs. deviant - /ba/; and standard - /pa/ vs. deviant - /p^ha /). They found that the phonemic contrasts were preserved for age-matched TD controls; however, the CAS group showed MMN responses to the phonetic contrasts only (allophonic variation, p^ha), with no evidence of an MMN response to the phonemic contrast. They concluded that CAS has a phonological component in addition to an output motor planning component.

1.5 Rationale for Dissertation Research

While there have been numerous studies evaluating the association of other idiopathic SSD and RD (Bird et al., 1995; Bishop & Adams, 1990; Cabbage et al., 2018; Larrivee & Catts, 1999; Nathan et al., 2004; Peterson et al., 2009; Raitano et al., 2004; Rvachew, 2007), few studies have examined this association for CAS (cf. Lewis et al., 2004; Miller et al., 2019; Stein et al., 2020). One explanation for the limited research is that CAS has been predominately conceptualized as a motor-speech disorder, limiting the pursuit of cognitive-linguistic research. Another reason is that CAS is a low-incidence disorder. Its prevalence in the general population ranges from 0.1% to 0.2%, or one to two children per thousand (Shriberg et al., 1997a; Shriberg et al., 2019) and is reportedly found in 2.0% to 2.4% of children referred for speech sound disorders (Shriberg et al., 2019). Research in communication disorders on low-incidence conditions such as CAS may not have access to large enough participant samples to conduct randomized controlled trials, the gold standard of research (Justice et al., 2008).

This dissertation research sought to contribute to our knowledge base about the association of RD and CAS by examining reading abilities in children with an early diagnosis of CAS and comparing them to children with a diagnosis of RD but with no history of SSD (RD-no SSD). This comparison may help identify the underlying skills (endophenotypes) associated with literacy for children with CAS relative to children with literacy deficits without an SSD. Identifying differing endophenotypes can guide appropriate treatment strategies and determine whether literacy-focused intervention should differ in these groups—a crucial consideration,

given the importance of aligning literacy intervention with a student's specific needs to be effective (Fuchs et al., 2017).

The study's first aim was to investigate the frequency of RD in a group of children with an early diagnosis of CAS. In light of previous research showing elevated rates of RD in children with CAS compared to children with other idiopathic SSD (Lewis et al., 2004; Miller et al., 2019), we hypothesized that children with an early diagnosis of CAS would demonstrate elevated rates of RD relative to other SSD and population estimates. We also hypothesized that the participants with CAS would demonstrate literacy deficits (i.e., word-level decoding impairment) similar to children with RD-no SSD. Given research reporting the deleterious effect that persistence of speech sound errors has on literacy beyond school entrance (Bird et al., 1995; Raitano et al., 2004), we expected that children with an early diagnosis of CAS who exhibited RD would demonstrate a similar level of impairment in decoding as children with RD-no SSD.

The second part of Aim 1 was to examine whether children with an early diagnosis of CAS would differ from children with RD-no SSD on the endophenotypes that have been identified to underlie literacy skills. We hypothesized that children with early diagnoses of CAS would demonstrate poorer phonological processing skills than children with RD-no SSD. Prior research has implicated a core phonological deficit affecting reading and speech sound production in CAS (Gillon & Moriarty, 2007; Marion et al., 1993; Marquardt et al., 2002; Miller et al., 2019). However, research has not demonstrated that a core phonological deficit is present in all cases of RD, especially for those without a prior SSD (Pennington et al., 2012; Peterson et al., 2009; Wolf & Bowers, 1999).

Language skills, including receptive vocabulary, were predicted to be decreased in both groups as many children with CAS and children with RD have been shown to have oral language deficits (Catts et al., 1999; Iuzzini-Seigel, 2019; Lewis et al., 2004; McArthur et al., 2000; Wise, 2007). However, we hypothesized that language skills would be poorer for the participants with an early diagnosis of CAS. Most studies report an increase in expressive language deficits for children with CAS relative to other forms of SSD (Ekelman & Aram, 1983; Lewis et al., 2004; Thoonen et al., 1997), and receptive language deficits have also been reported (Lewis et al., 2004). While studies have demonstrated that children with RD (Ziegler et al., 2009) and children with CAS have difficulties with speech production in noise (Iuzzini-Seigel, 2015), speech-in-noise perception was predicted to be poorer for the CAS participant group. Studies have consistently shown that speech perception underlies difficulties for children with a diagnosis of SSD, including CAS. Motor-speech skills were predicted to be poorer in the participants with an early diagnosis of CAS than children with RD-no SSD; difficulties with motor-speech sequencing skills are considered a hallmark of CAS (Peter et al., 2012). However, previous studies have shown that children with RD also have difficulty with multi-syllable word repetition (Catts, 1986) and rapidly sequencing single syllables (Fawcett & Nicolson, 2002).

The study's second aim was to determine if there were subgroups within the CAS cohort that could be differentiated by reading ability. There is evidence that children with an early diagnosis of CAS can exhibit average and below-average decoding (Miller et al., 2019; Stein et al., 2020). We hypothesized that subgroups within the CAS group would be distinguished by average or below-average performance in word-level decoding fluency. Furthermore, we

hypothesized that decoding ability would be associated with PA skills, speech sound production, and speech in noise perception. Children with RD have demonstrated deficits in PA skills (Gillon & Moriarty, 2007; Marion et al., 1993; Marquardt et al., 2002; Miller et al., 2019), and speech in noise perception (Ziegler, 2009). These same deficits are hypothesized to underlie literacy skills for children with CAS. The association of persistent speech sound production errors and decoding has been equivocal, with some research showing an association between poorer speech production and reading (Lewis et al., 2004) and others showing limited association (Miller et al., 2019, Stein et al., 2020). Although our sample size of participants with an early diagnosis of CAS was small, our results may serve as preliminary data for future investigations.

Chapter 2. Historical Roots and Current Conceptualizations of CAS

2.1 History of CAS

In his opening remarks at the 2002 Childhood Apraxia of Speech Research Symposium, Joseph Duffy quoted Carl Sagan, "When knowledge is lacking, a name comes to take its place" (Duffy, 2002, p. 7). Accordingly, a significant lack of knowledge about CAS[2] can be observed by proxy in the many names that researchers have used to describe it. There are as many as 21 labels that have been applied to this disorder since 1937 (Hall et al., 2006). Some of the more common ones include Developmental (Dys)Apraxia (Orton, 1989; McCabe et al., 1998); Articulatory Dyspraxia (Morley et al., 1954); Developmental Articulatory Dyspraxia (Morley, 1957); Developmental Apraxia of Speech (Hall et al., 1993, 2006); Developmental Apraxias of Speech (Crary, 1993); Developmental Verbal Dyspraxia (Aram & Nation, 1982; Velleman & Strand, 1994); Suspected Developmental Apraxia of Speech (Shriberg et al., 1997a, 1997b), and the latest iteration, Childhood Apraxia of Speech (ASHA, 2007). These terms reflect different conceptualizations of the disorder including, a motor-speech disorder with co-occurring language and literacy issues, a motor-speech disorder with a core linguistic deficit, and a syndrome or symptom complex with associated deficits arising from a heterogeneous genetic source (Chilosi et al., 2015; Worthey et al., 2013).

[2] Childhood Apraxia of Speech (CAS) will be used herein to refer to all historical references of pediatric apraxia of speech. In addition, CAS herein refers to the "idiopathic" disorder as distinguished from CAS associated with a known neurogenic impairment (ASHA, 2007).

2.1.1 Apraxia of Speech in Adults

Conceptualizations of CAS have been partially derived from the adult form of the disorder, Apraxia of Speech or AOS, which emphasized the functional modularity between speech and language deficits in adults who have suffered brain damage (Darley, 1967; Wepman & Van Pelt, 1955). However, the idea that a speech deficit could exist separate from a language deficit has been controversial from the outset. The roots of this historical controversy, traced below, extend into the present for both AOS and CAS and inform current thinking about these disorders.

The word apraxia is Greek in origin and means without action (Pearce, 2009). In 1870, Steinthal, a German linguist, described individuals with aphasia (an acquired or neurodegenerative language disorder) who were unable to perform sequences of movement such as the use of familiar objects (e.g., a comb) as having apraxia, despite retaining adequate strength and coordination (Pearce, 2009). However, Liepmann, a German neurologist, provided the "historically dominant and widely accepted conceptualization of apraxia" (Duffy, 2013, p. 272). Liepmann established subtypes of apraxia such as ideo-motor apraxia—the inability to follow verbal commands or mimic an action such as waving goodbye; limb-kinetic apraxia—clumsiness in performing a precision, not due to paralysis, muscle weakness, or sensory loss; and ideational apraxia—an inability to use familiar objects upon command appropriately.

While Liepmann proposed apraxia as a class of general movement disorders, he did not specify an *apraxia of speech* (McNeil, 2002). Liepmann believed that defects in speech were most often the result of aphasia. However, in certain prescribed circumstances, it could be an

apraxia. He used the term *motor aphasia* to describe a special form of apraxia of the "glosso-labio-pharyngeal musculature: the nonparalyzed muscles cannot be innervated to bring forth the sounds of speech" (Brown & Liepmann, 1988, p. 34). Liepmann's concept of motor aphasia was consistent with Broca's earlier use of the term *aphemia*, which he used to describe a patient in 1861 with a "disturbance in the organization of articulatory and motor aspects of speech" (Fox et al., 2001, p. 123). Broca observed:

> There are cases in which the general faculty for language remains unaltered; where the auditory apparatus is intact; where all muscles—including those of speech and articulation—are under voluntary control; and where, nevertheless, a cerebral lesion abolishes articulated language. This abolition of speech, for individuals who are neither paralyzed nor idiots, constitutes such a singular symptom that it appears useful to designate it under a special name. I will therefore give it the name of aphemia. (cited in Henderson, 1990 p. 85)

However, by 1869, Broca recognized that the explicit articulatory disorder of aphemia was rare and that most often it was accompanied by difficulties in oral comprehension, reading, and writing (Henderson, 1990).

The proposition that an impairment of speech could exist in the absence of language deficits post-CVA (cerebrovascular accident) was advanced in the 1950s by Wepman, a noted aphasiologist. He proposed two distinct disorders, "symbolic or integrative," consistent with aphasia, and "non-symbolic or transmissive," consistent with apraxia or agnosia (Wepman &

Van Pelt, 1955, p. 234). Wepman's conceptualizations of apraxia-like speech behaviors fueled a renewed interest in assigning different labels for putatively distinct behaviors. Darley, a speech-language pathologist, and researcher, echoed Wepman in observing that motor aphasia had "no cross-modality impairment in the use of language symbols but a specific modality-bound deficit, better labeled an apraxia" (Darley, 1967, p. 236). In a 1969 presentation to an ASHA convention in Chicago, Darley explicitly outlined his conceptualization of apraxia of speech (AOS).

> An articulatory disorder resulting from impairment, as a result of brain damage, of the capacity to program the positioning of speech musculature and the sequencing of muscle movements for the volitional production of phonemes. No significant weakness, slowness, or incoordination in reflex and automatic acts. Prosodic alterations may be associated with the articulatory problem, perhaps in compensation for it. (cited in Rosenbek, 2001, p. 270)

Darley's formal announcement of AOS as a motor-speech disorder entirely separable from aphasia initiated a contentious debate. Many aphasiologists saw AOS as a set of symptoms consistent with aphasia "that can lend themselves equally well to a theory of interrelationships with other processes of language as opposed to a theory of discrete motor impairment'' (Martin, 1974, p. 63). The debate focused on speech symptoms. For example, phonemic paraphasias, sound substitutions, and mis-sequenced sounds resulting in words that retain some semblance of the intended word, are often evident in post-CVA speech output. Examples include anticipatory errors—apple > papple, perseverative errors—gingerbread > gingerjed, and transposition errors —pneumonia > menonia. Those supporting the diagnostic label of AOS argued that the

paraphasic errors in individuals with AOS were random and not related linguistically to the original phoneme (Aten et al., 1975). However, aphasiologists who opposed the concept of AOS believed that these same examples were associated with the original word and not random. Furthermore, they thought that these errors were due to phonological encoding errors (i.e., phoneme selection and sequencing) occurring at the stage before a word is transformed into a motor plan. Martin (1974) argued the more appropriate name for AOS symptomatology was "aphasic phonological impairment," thereby removing bias and "identif[ying] the complex under discussion without connotations of a basic motor or perceptual disorder" (p. 63).

Difficulty in producing longer, more complex multi-syllabic *real* words was also cited as evidence of the motoric nature of AOS speech issues (Johns & Darley, 1970). However, opponents challenged this assertion and pointed out that aphasic individuals also had difficulty repeating real words of increasing length, and similar to subjects with AOS, they were also more successful in repeating real words than nonwords. Language experience had a definitive impact on speech production in aphasia and in AOS also (Martin, 1975).

A similar debate surfaced about the reported absence of auditory-perceptual difficulties associated with AOS. For example, individuals described as having AOS were said to have intact auditory perceptual abilities—"Apraxia of speech may occur in a fairly pure form without sensory, or at least without auditory components" (Aten et al., 1971, p. 142). Those who opposed the AOS label were quick to point out that the tasks used to assess auditory discrimination and sequencing in AOS patients were verbal to visual tasks, not characteristic of online language processing (i.e., the patient heard a series of words and then pointed to pictures in the correct

serial order) (Martin, 1974). Finally, it was made clear that an oral apraxia, such as the inability to perform oral tasks like sticking out the tongue or pursing the lips on command, may coexist with AOS; however, it may present itself entirely separately (Darley et al., 1975).

2.1.2 Apraxia of Speech in Children

While the debate about AOS was evolving, researchers became interested in investigating the motor-speech issues in pediatric speech and language disorders. Much of the interest was generated from the same individuals who were attempting to establish the basis for AOS. According to Duffy (2002), "They brought a combination of bias and valuable insight to their efforts" (p. 7). However, several researchers had investigated motor-speech disorders in pediatric populations much earlier, separate from the debate about AOS. For example, in 1891, Hadden, considered the "Broca of childhood AOS" (Duffy, 2002, p. 6), published, along with two other British physicians, articles describing boys who had "defects of articulation" that were so severe that their speech output was referred to as *idioglossia* because it sounded as if they were speaking a made-up language (Lorch & Hellal, 2012).

Orton, a neurologist who specialized in developmental dyslexia, was one of the earliest researchers (during the 1920s) to link speech sound production, reading, and general body coordination development in children. He used the term *developmental apraxia* to describe children who were "abnormally clumsy" with trouble "carrying-out of any complex trained movements," including "speech and writing" (in Orton, 1989, p. 72). Orton observed the connection between learning complex motor movements and their simultaneous effect on speech, language, and written language abilities. He postulated that the remediation for children

diagnosed with developmental apraxia revolved around "sequence building," theorizing that the temporal and spatial sequencing required for fluent speaking and reading was undermined by a lack of cerebral dominance (in Orton, 1989, p. 317).

Nearly three decades later, Morley, a British pediatric speech-language pathologist, used the term *articulatory apraxia* to distinguish it from developmental dysarthria (Morley et al., 1954). She noted that children diagnosed with articulatory apraxia had articulation difficulties but retained normal voluntary movements of the tongue, lips, and palate. Additionally, Morley noted that there were often accompanying delays in language and reading (Morley et al., 1954). Later, the diagnostic label was changed to *developmental articulatory dyspraxia* to describe articulation that was "inadequate for the complex and rapid movements involved in normal conversational speech" (Morley, 1957, p. 217). Morley documented the literacy outcomes for 11 children and one young adult diagnosed with developmental articulatory dyspraxia, whom she followed into school-age and beyond. She observed that of the 10 study participants followed to literacy acquisition, three were diagnosed with dyslexia and one with a spelling disability (Morley, 1957). Morley observed that children with developmental articulatory apraxia could be slow in learning to read and that the dyslexia associated with the disorder was not a secondary consequence (Morley, 1959).

Despite Morley's use of *developmental articulatory apraxia*, the preferred term for severe speech production difficulties during the 1950s and 1960s was expressive aphasia. The term aphasia was used to describe childhood language disorders that "mainly [fell] into two general types: those who cannot understand what others say and those who cannot speak"

(Myklebust, 1961, p. 7). Consistent with Myklebust, Wilson (1965) recognized two forms of childhood aphasia: Class I, "congenital motor or expressive aphasia," and Class II, "congenital sensory or receptive aphasia" (p. 10). Class I motor/expressive aphasia criteria were normal intelligence, hearing, and language comprehension, coupled with an inability to imitate words and imitate speech sounds (McGinnis, 1963). Additionally, oral-facial apraxia could be present; therefore, when diagnosing motor aphasia, "[testing] requires the child to imitate lip and tongue actions" to assess accompanying oral apraxia (Wilson, 1965, p. 10).

Rosenbek and Wertz (1972) examined the differences between AOS and a developmental form of the disorder and advocated for use of the term *apraxia* over expressive aphasia for children.

> Since Wilson's description better fits an articulatory or motor speech disorder rather than a central language disorder, such a child might more correctly be said to have an apraxia of speech or a developmental apraxia of speech . . . The term apraxia, by emphasizing the articulatory or motor programming deficit in these children, differentiates them from children with a central language impairment—aphasia—and suggests efficacious therapeutic procedures. (p. 23)

They investigated the speech and language characteristics of 50 children with motor speech difficulties. A significant finding of the study was the number of children with general body apraxia and oral apraxia. EEGs were available for 26 children with evidence of right hemisphere involvement. The authors suggested that praxis (i.e., ability to perform skilled movements of

speech) in children involves large cortical areas of both hemispheres, unlike the discrete left-hemisphere lesions in Broca's area involved in AOS. Vowel errors, groping, trial and error behavior, errors on longer stimuli, and evidence of oral apraxia differentiated a child with developmental apraxia from a child with a functional articulation disorder. Notably, 56% of the children in the study had an accompanying language disorder (aphasia). The authors cautioned that the incidence of *isolated* developmental apraxia (no accompanying language disorder) in their sample (18%) might have been

> spuriously high . . . influenced by the difficulty in measuring aphasic involvement in children, both because the behaviors to be called aphasia in children are not agreed upon by all speech pathologists and because language testing is difficult in children with severe output disturbances. (Rosenbek & Wertz, 1972, p. 26)

Yoss and Darley (1974), also hoping to extend an understanding of AOS versus a developmental form of apraxia, investigated articulatory deficits in children to determine if a pediatric population would demonstrate "behavior evident in some brain-injured adults" (p. 399). Sixty children, ages 5 through 10 years (30 age-matched controls, with no history of speech difficulties, and 30 children in a group labeled "defective articulation children" or DAC), were evaluated. Children were assessed on the following variables: auditory perception and discrimination, execution of volitional oral movements, phoneme production in spontaneous contextual speech, phoneme production in real and nonsense words, and oral diadochokinetic rate. Unlike Rosenbek and Wertz (1972), participants in the Yoss and Darley study were excluded if language abilities were not within the normal range. However, language status was

ascertained by a screening test, the Utah Test of Language Development (Mecham et al., 1973), which may not have been adequate to assess language abilities for children with significant speech issues.

As expected, differences surfaced between the DAC and control group in the number of phoneme errors, the production rate of individual syllables in the diadochokinetic tasks, and the repetition of correct syllable order. The Denver Auditory Phonemic Sequencing Test (DAPST) (Aten, 1973) was used to assess auditory perception and discrimination skills. Children heard a series of words, from two to six items, with minimal phonemic variance. Participants had to retain the information and recognize the sequence visually. Significant differences surfaced between the groups, with the DAC group scoring significantly lower than the controls, signifying an association of deficits in perceptual skills and speech production deficits.

An additional goal of the study was to determine if there were subgroups within the DAC group. Significant group differences surfaced in "soft" neurologic signs and performance on an oral movement task, with the DAC group displaying evidence of neurologic symptoms and oral movement difficulties. The two groups also differed by speech errors, with the apraxic group exhibiting multiple feature errors, distortions, prolongations and repetitions, additions, and omissions of sounds. The authors concluded that they had identified "a cluster of . . . children . . . whose group performance lends substantial support to the use of the term developmental apraxia of speech" (Yoss & Darley, 1974, p. 412).

Several other key findings emerged from the study. The speech characteristics of adults with AOS appeared to differ from children. Groping and searching for the correct placement of the articulators, characteristic of adults with AOS, was not typical of children, who demonstrated no awareness of their errors. Furthermore, oral-motor apraxia was often comorbid with children but not adults.

Williams et al. (1981) attempted to replicate Yoss and Darley's (1974) findings. The replication study was prompted by Williams' unsuccessful attempts to use the diagnostic criteria established by Yoss and Darley to clinically diagnose children with developmental apraxia of speech. A range of articulatory problems was identified but none delineated a subgroup beyond the larger group status of functional defective articulation. The replication research appeared paramount at the time as

> Yoss and Darley's findings have been used to uphold the notion that there exists a subgroup of defective articulation children called dyspraxic. Consequently, their findings have supported the development of alternative treatment strategies for this dyspraxic subgroup. At the very least, the present study's failure to support Yoss and Darley's findings should raise questions about the premises on which this clinical literature has grown—and is growing" (p. 503)

Other researchers also questioned the existence of CAS as a clinical entity. Guyette and Diedrich (1981) surmised from their literature review that "developmental apraxia of speech is a label in search of a population" (p. 39). Over the next several decades, the debate continued with

motor speech and linguistic components of CAS being variously emphasized. Developmental verbal dyspraxia (DVD), a term advanced by Aram (1984), reflected her concept of the disorder as a "syndrome complex." Aram described DVD as being "as much language-based as articulatory , , , [with] all levels of formulated speech affected . . . involving reading and written expression" (Aram & Nation, 1982, p. 161). Velleman and Strand (1994) also used DVD to describe CAS as a "disorder of hierarchical organization," which affected both linguistic and motor-speech movements (p. 120). Crary (1993) coined the term *developmental apraxias of speech* and proposed a "motolinguistic" continuum of clinical symptoms resulting from overlapping functions along the "anterior-posterior dimension" of the left hemisphere (p. 60). Crary also described motor-speech disorders as disorders with "a high incidence of related learning disabilities—especially difficulty with reading" (p. 28).

For Hall et al. (1993), the term *developmental apraxia of speech* (DAS) represented a view of the disorder as solely one of speech motor control, related to speech production. DAS seemed the most appropriate diagnostic label as the basis for the problem was one of motor-programming for speech, not verbal or linguistic. Language problems were viewed as concomitant, part of the myriad of problems that co-occurred in children with DAS. Language issues seemed to improve quickly as speech issues resolved; seemed indicative of the "co-occurring" nature of linguistic difficulties rather than "the entirety of the disorder" (p. 25).

Ozanne (1995) advanced a view of CAS antithetical to Hall et al. (1993), conceptualizing CAS as a multi-deficit disorder. She posited that CAS involved three levels of impairment—a phonological planning deficit, a phonetic assembly impairment, and a motor-program execution

impairment. “Flow on effect” and “feedback effect” from these three levels could cause deficits in a top-down manner or from the lower level upwards (p. 106). For example, deficits at one level can cause difficulties at ascending and descending levels (e.g., early motor deficits cause phonological processing difficulties).

2.2 Current Perspectives of CAS

In the second edition of *Developmental Apraxia of Speech*, Hall et al. (2006) continued to conclude that DAS was a disorder of “speech-motor control with the focus on speech”; language disorders, academic difficulties, gross and fine motor difficulties, and feeding problems were considered co-occurring with evidence lacking to link “those other issues to the speech disorder itself” (p. 11). In 2007, ASHA released its technical report outlining a position statement on CAS. It contained a motor-speech orientation similar to Hall et al. (2006). CAS was defined as “a speech disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits, e.g., abnormal reflexes, abnormal tone” (ASHA, 2007, p. 3). ASHA acknowledged the presence of additional difficulties in CAS, considered “co-occurring characteristics or symptoms,” of delayed language development; expressive language problems such as word order confusion and grammatical errors; problems learning to read, spell, and write; and problems with social language/pragmatics. (ASHA, n.d.).

The ASHA Technical Report (2007) specifies that “at present, there is no validated list of diagnostic features of CAS that differentiates this symptom complex from other types of childhood speech sound disorders, including those primarily due to phonological-level delay or neuromuscular disorder (dysarthria)” (p. 5). However, ASHA (2007) determined three features

of the disorder to have gained consensus for diagnosis and are currently in use as the gold standard of a CAS diagnosis:

(1) inconsistent errors on consonants and vowels in repeated productions of syllables or words

(2) lengthened and disrupted coarticulatory transitions between sounds and syllables

(3) inappropriate prosody, especially in the realization of lexical or phrasal stress (p. 3)

Despite an apparent consensus regarding the nomenclature and diagnostic variables for CAS, a lack of agreement about how to define the disorder continues. Nijland et al. (2015) examined the role of complex sensorimotor and sequential memory functions (e.g., number recall, imitating a sequence of hand movements, etc.), and simple sensorimotor functions (e.g., tapping with index fingers, identifying finger localization, etc.), in a group of typically developing children and children with CAS on two occasions separated by 15 months. The participants with CAS performed more poorly on all tasks on both occasions and demonstrated disordered complex sensorimotor and sequential memory functions. Nijland et al. (2015) identified two conceptualizations of CAS:

> (a) CAS is a unitary disorder, most likely a disorder of sequencing speech movements, with a nonverbal sequential comorbidity in most children with CAS. (b) CAS is a symptom complex, primarily comprising errors of sequencing at diverse levels of speech movements (segmental, syllabic, suprasegmental); there is no single, common underlying deficit for all children with CAS, but the symptom complex can be the result of different, specific underlying deficits, followed by different possible developmental trajectories. (p. 559)

Based on their findings, Nijland and colleagues determined that CAS was a “symptom complex” involving difficulties in both sequencing of speech movements and in nonverbal sequential cognitive abilities.

Peter et al. (2013) also investigated the hypothesis of a global deficit in sequential processing in a multigenerational family with a history of CAS. Compared to unaffected members, affected family members demonstrated difficulties with multisyllable real and non-word imitation, decoding of real and nonsense words, and spelling. Consistent with the findings of Nijland and colleagues, they proposed that CAS involves a global deficit in sequential processing, affecting speech, cognitive and linguistic tasks and concluded that CAS was not a unitary disorder of motor speech sequencing with comorbidities.

In a more recent study, Peter et al. (2018) investigated whether individuals with dyslexia and individuals with CAS share a common global deficit in processing complex sequential information. Participants were adults with a history of CAS, adults diagnosed with dyslexia, and neurotypical controls. All participants were assessed for real and nonword multisyllable repetition, real and nonword decoding, and diadochokinetic rate. Responses were analyzed for phonological processes involving two classes of errors, sequencing errors and substitution errors. Despite the different diagnoses, RD versus CAS, analyses of verbal error patterns revealed that both disorder groups showed a persisting deficit in sequencing errors (sequential processing). Peter et al. (2018) suggested that the similarity in phenotype may be the result of a "shared deficit in cerebellar function caused by genetic variations" (p. 339).

Iuzzini-Seigel (2019) investigated speech, language, and gross and fine motor impairment

for children with SSD, including CAS. Significant associations surfaced between the number of CAS speech features and deficits in motor tasks (i.e., manual dexterity, balance, and aiming and catching) and performance on the GFTA-3 and motor tasks, especially for those children with accompanying language impairment. Iuzzini-Seigel suggested that a higher-order deficit, a *procedural learning deficit,* leads to co-occurring speech, language, cognitive, and motor impairments for some children with CAS. The procedural learning system is hypothesized to regulate how children learn "patterns (e.g., phonological patterns, grammatical rules) without being explicitly taught" (p. 3222).

In another study of procedural learning, Iuzzini-Seigel (2021) investigated whether children with CAS, children with other idiopathic SSD, and typically developing children would perform differently on procedural learning involving serial reaction time. While there was a small subgroup of low performers in the SSD and TD groups, children with CAS generally required an increased number of exposures to a visuospatial sequence to demonstrate procedural learning. This may help to explain why children with CAS need increased frequency and intensity of intervention to learn speech targets. Iuzzini-Seigel's findings support a "global sequencing deficit framework" in CAS, similar to Peter et al. (2013) and Nijland et al. (2015).

There has also been genetic research on CAS. Current trends in genetic research support a widespread locus for CAS. For example, in a genetic study of two multigenerational families with histories of SSD consistent with CAS, evidence was found to support a CAS phenotype similar to other neurological phenotypes such as Alzheimer's disease and autism spectrum disorder (Peter et al., 2016). The authors state that CAS is a "complex and heterogeneous

disorder with several discoverable variants, each of which segregates and confers risks of varying levels of impact" (p. 20). In another genetic study of 10 unrelated participants with CAS, three of the 10 participants expressed gene, KIAA0319, associated with developmental dyslexia and SLI (Worthey et al., 2013). Like other complex neurodevelopmental disorders, the authors concluded that CAS has heterogeneous genetic origins.

Liégeois et al. (2019) investigated the structural and functional impairments underlying CAS using MRI brain scanning and genetic analysis. Although no significant genetic findings surfaced, they found reductions in the axonal integrity bilaterally in the arcuate fasciculus and reductions in the left temporoparietal cortex, indicating a developmental disorder of the dorsal language route. The dorsal route is proposed to link "auditory/input representations to articulatory systems and transform phonological representations into motor programs" (Liégeois et al., 2019, p. 973). This same pathway is proposed as the functional and neuroanatomical locus of the DIVA model's auditory feedback system, "in particular, the arcuate fasciculus" (Guenther, 2016, p. 164). Liégeois et al. (2019) proposed that during the babbling stage of speech development, the dorsal route, with its initially underdeveloped connections between the auditory and premotor cortex, "undergo[es] progressive tuning" as typically developing infants create connections between auditory input regions and the motor cortex (p. 974). If these connections do not develop properly during the initial stages of an infant's speech and language development (babbling and imitation), the transformation of phonological representations to motor plans may be compromised.

Hildebrand et al. (2020) performed precise phenotyping and research genome or exome

analysis on 34 children with a primary diagnosis of CAS. Results supported the idea that CAS is "highly genetically heterogeneous, often occurring as a sporadic monogenic disorder" (p. 2166). The phenotyping data suggested that CAS is often not an isolated speech impairment but more closely resembles a wide-ranging neurodevelopmental disorder as the participants (n = 34) had a range of deficits including motor skills, cognition, attention, behavior, emotional regulation, toileting, and social skills. Hildebrand et al. found no apparent differences between the phenotypes of children whose molecular genetic diagnoses had been confirmed compared with those with uncertain or no genetic findings.

2.3 Summary

Reflecting on the past, Ziegler (2012), described Darley's research as the origin of much of the current definitional and diagnostic criteria of AOS. However, Ziegler pointed to ongoing "uncertainties" in the current conceptualizations of AOS reflecting

> a fundamental under-specification of our models describing how stored word form representations are transformed into actual speech movements, whether and how this process can be subdivided into substages, how the alleged substages and their boundaries are represented in the brain, and—as a consequence—how their clinical correlates can be delineated. (p. 1498)

If the anatomical and functional interrelationships of the modules of phonological encoding, motor planning, and execution are not delineated in an adult neurogenic disorder such as AOS, i.e., the "alleged substages and their boundaries in the brain," they are even less discernable in a developmental disorder such as CAS. As noted by Karmiloff-Smith (2006), "modules could be

the result of ontogenesis over developmental time, not its starting point" (p. 11). Furthermore, "even a tiny asynchrony or impairment early on in development can have a huge, cascading impact on the phenotypic outcome" (p. 15).

The behavioral, genetic, functional imaging, and neurocomputational studies reviewed above support a view of CAS as a complex, heterogeneous disorder with a pathognomonic motor speech component accompanied by a complex set of associated deficits (Chilosi et al., 2015; McCabe et al., 1998; Worthey et al., 2013). CAS is not a unitary disorder with children demonstrating the same set of speech symptoms (Maassen, 2015; Nijland et al., 2015; Velleman, 2016); instead, children can evidence differing levels of severity of motor speech involvement, differing speech characteristics, and speech characteristics that change in type and frequency as children mature. Furthermore, 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). Accordingly, language and reading difficulties can be considered part of the disorder's "symptom complex" (Nijland et al., 2015; Velleman, 2016). Maassen (2015) advocates for a developmental perspective of CAS as a disorder that is not dissociated from language acquisition but a developmental motor speech disorder that impacts other domains such as "phonological and lexical development and auditory–perceptual functions" (p. 131).

Chapter 3. Methodology

This study used a standard two-group comparison cross-sectional design to assess word-level decoding skills and the underlying differences in cognitive and speech-language skills related to decoding skills in two independent groups of school-age children and adolescents.

3.1 Participants

Participants were school-age children and adolescents ranging in age from 8 to 14 years with an early diagnosis of a CAS (n = 16) or a diagnosis of RD-no SSD (n = 16). Participants with an early diagnosis of CAS were recruited from the caseloads of private speech-language pathologists and by posting information about the research study on known CAS parent advocacy groups. Participants with RD-no SSD were recruited from the caseloads of reading specialists, learning center teachers, private schools specializing in learning differences, and by posting information about the research study on websites of known parent advocacy groups for RD.

Due to COVID-19, all in-person research at Case Western Reserve University was suspended in March of 2020. Additional participants were required to create adequate power for the study. Three school-age children (M = 10;4 years) with an early CAS diagnosis who had undergone evaluation in a concurrent research study of SSD, i.e., Biomarkers of SSD, were selected based on age. All three participants met the same general inclusion and criteria for confirmation of an early diagnosis of CAS as outlined below and had undergone a similar battery of assessments. The participants' parents were contacted for permission to utilize their child's previously collected data, as the Case Western Reserve IRB required.

3.2 Procedures

Once apprised of the research study, parents interested in having their child participate contacted the Principal Investigator (PI). The PI responded to all parents' inquiries to participate in the study, following a two-step screening process. The first step was a phone interview during which the following general inclusion requirements for the participant were established: 8 to14 years of age, monolingual English speaker, absence of a history of neurological disorders other than CAS or RD (e.g., cerebral palsy or autism spectrum disorder), reported IQ in the normal range, and a reported diagnosis of CAS or RD-no SSD. Once general inclusion requirements were established, an appointment was made for the child, accompanied by their parent(s), to come in for an in-person assessment.

Testing was conducted on a single day at Case Western Reserve University at the Cleveland Hearing and Speech Center in a quiet office on the fourth floor. At the parent's request, testing was also performed in a quiet and adequately lit room in the family's home (n = 4). Testing was conducted individually in two sessions of approximately 2.5 hours each, with a break for lunch. The PI, a licensed, ASHA-certified speech-language pathologist and reading specialist with experience in assessing children with SSD and decoding difficulties, performed all evaluations. Initial responses were transcribed while the child was speaking using broad phonetic transcription and audio recorded for later narrow transcription. The Institutional Review Board of Case Western Reserve University of Cleveland approved the study, and informed consent and assent were obtained from all participants prior to the start of testing. See below for a description of the General Inclusion Measures (also shown in Table 1, General Inclusion Measures).

3.3 Measures

3.3.1 General Inclusion Measures

The following is a description of the measures used to determine general inclusion criteria for all participants in the study:

The Kaufman Brief Intelligence Test-2nd Edition (KBIT-2) (Kaufman & Kaufman 2004) was administered to assess non-verbal, performance IQ (PIQ). Only participants with a standard PIQ score of 80 or above were included in the study.

Oral Speech Mechanism Screening Examination-3rd Edition (OSMSE-3, St. Louis & Ruscello, 2000) was administered to rule out frank oral structural anomalies and to evaluate muscle weakness associated with dysarthria, which is often comorbid with CAS.

Hearing Screening was conducted to ensure normal hearing acuity. All children passed a hearing screening at 20 dB HL at all octave frequencies between 250 and 8000 Hz, bilaterally. Hearing testing was conducted with a portable audiometer (a Beltone 119) using Telephonics Dynamic Headphones (TDH) with protective, disposable earphone booties.

Table 1

General Inclusion Measures

Domain	Measurement	Purpose	Requirements	Values Reported
Cognitive	The Kaufman Brief Intelligence Test-2nd Ed. (KBIT-2)	To assess non-verbal IQ (PIQ)	Standard PIQ score $\geq$ 80	Standard scores
Oral Structure	Oral Speech Mechanism Screening Examination - 3rd Ed. (OSMSE-3)	To document normal structure and assess muscle strength and tone	Must demonstrate normal structure	Pass/Fail
Developmental/ Background Information	An Initial Phone Interview	To ascertain developmental information	Between the ages of 8 and 14 years, monolingual English speaker, with no history of neurological disorders (e.g., autism spectrum disorder, cerebral palsy), diagnosis of CAS or RD-no SSD	Parent Report
Hearing	Audiometric Hearing Screening	To ensure hearing acuity at 20 dB HL at octave frequencies of 250-8000 Hz, bilaterally	Demonstrate normal hearing acuity	Pass/Fail

3.3.2 Documentation of a CAS or RD-no SSD Diagnosis

The study included school-age children and adolescents with a *documented* history of a CAS or RD diagnosis. With regard to the CAS participants, the PI, a licensed speech-language pathologist, confirmed an early diagnosis of CAS by using at least one of the following criteria: presentation of a previous or current diagnostic evaluation performed by a licensed SLP or neuropsychologist stating the child had a CAS diagnosis, a current or past Individualized Educational Plan (IEP) with a statement regarding a CAS diagnosis, or a detailed description of a CAS diagnosis, including dates of diagnosis and a description of ongoing or past speech-

language therapy on a Developmental Questionnaire (Appendix C). The three participants recruited from the previous study of SSD were confirmed to have an early diagnosis of CAS by an ASHA certified SLP serving on the study's research team during an initial evaluation in preschool. Herein, to simplify description, participants with an early diagnosis of CAS will be referred to as *participants with CAS* or *participants with a diagnosis of CAS* or *CAS diagnosis*.

The PI confirmed the diagnosis of RD-no SSD using at least one of the following criteria: presentation of a previous diagnostic evaluation report performed by a qualified reading specialist or neuropsychologist, an IEP with a statement regarding an RD diagnosis, and a detailed description of ongoing concerns about RD, or a detailed description of RD on the Developmental Questionnaire, including dates of diagnosis and description of reading intervention. For RD participants, the absence of any history of an SSD was confirmed by the initial telephone screening, an in-depth discussion with a parent before the start of testing, review of records, and observation of the participant's articulation during the evaluation.

The Hollingshead Four Factor Index of Socioeconomic Status (Hollingshead, 1975) is a survey designed to rate an individual's social status based on four domains: marital status, employment status, educational attainment, and occupational prestige. A socioeconomic status score was calculated for each participant's family based on information provided in the questionnaire.

3.3.3 Experimental Measures for the CAS and RD-no SSD Groups

The following is a description of the experimental measures administered to all participants in the study. Additional information pertaining to the experimental measures and their respective domains and prediction for each assessment is provided in Table 2.

The Peabody Picture Vocabulary Test-Fifth Edition (PPVT-5) (Dunn, 2018) is a norm-referenced assessment of single-word, receptive vocabulary. It provides a broad measure of oral vocabulary and verbal ability in a point to picture format in response to a heard word. Smaller vocabulary size has been cited as a consequence of both SSD and RD but with differing explanations (Cunningham & Stanovich, 2001; Stanovich, 1993; Stanovich & Cunningham, 1993; Stoel-Gammon, 2011). Scores used in analysis were age-adjusted standard scores with a mean of 100 and a standard deviation of 15. Scores between 85 and 115 are considered within the "expected" range.

The Clinical Evaluation of Language Fundamentals-Fifth Edition (CELF-5) (Wiig, Semel, & Secord, 2013) is a standardized measure of receptive and expressive language abilities and was administered to evaluate the general language abilities in the participant sample. The Core Language Score includes four subtests from the larger CELF-5 battery that best discriminate typical versus disordered language performance. Scores used in analysis were age-adjusted standard scores with a mean of 100 and a standard deviation of 15. Standard scores between 86 and 114 are within the average range.

The Comprehensive Test of Phonological Processing-Second Edition (CTOPP-2) (Wagner et al., 2013) measures the construct of phonological processing, a set of cognitive-linguistic skills thought to underlie both speech and reading development. Phonological processing comprises three components: PA, PM, and RAN. The CTOPP-2 was administered to determine the strengths and weaknesses in a participant's phonological processing skills and examine the association of these skills with word-level decoding proficiency.

Two composite scores (i.e., Phonological Memory and Rapid Symbolic Naming) and two subtests that comprise the Phonological Awareness Composite (i.e., Elision and Word Blending) were used in the analysis. The composite scores are standardized scores with a mean of 100 and a standard deviation of 15. The Elision and Word Blending subtests use a scaled score with a mean of 10 and a standard deviation of 3. Scaled scores were converted to standard scores. Standard scores of 90 to 110 are within the average range.

Three participants with CAS taken from the Biomarkers of SSD study did not have subtest data for the Phonological Memory Composite and were missing one subtest used to form the Phonological Awareness Composite (Phoneme Identification). As a result, the Phonological Memory Composite score was based on 13 participants with CAS. The Elision and Word Blending subtest scores were used in the analyses in lieu of the Phonological Awareness Composite as they included all 16 CAS participants.

The Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate (Fletcher, 1978) was used to measure motor-speech sequencing skills. Deficits in motor speech sequencing are a hallmark in the diagnosis of CAS (Rvachew & Brosseau-Lapré, 2018; Thoonen et al., 1999). Motor-

speech sequencing skills are also associated with RD in children without SSD (Fawcett & Nicolson, 2002). This assessment evaluates a child's ability to rapidly repeat single syllables (pʌ, tʌ, kʌ, fʌ, lʌ), bisyllables (pʌtə, pʌkə, tʌkə) and multisyllabic sequences (pʌtəkə). Participants were asked to produce five monosyllabic syllable repetitions, three, two-syllable repetitions, and one, multisyllabic repetition. The mean score from all of these tasks was entered into analysis. Scores used in analysis were age-adjusted standard scores based on normative standards through 13;11 years.

Multisyllabic Word Repetition (adapted from Catts, 1986) was used to assess speech production in real, complex words. This task assesses access to lexical information and the retrieval of accompanying motor plans in multisyllable words. Participants were asked to repeat a list of 20 words after the examiner, and the responses were phonetically transcribed and analyzed for phonological processes and syllable structure. The percentage of correctly produced words was calculated and transformed into z-scores based on the performance of unaffected siblings of probands (n = 548) assessed as part of a larger longitudinal study, The Cleveland Family Speech and Reading Study. These scores were transformed into standard scores with a mean of 100 and a standard deviation of 15 to be comparable to the other variables in the study and entered into analysis.

Nonsense Word Repetition (adapted from Kamhi & Catts, 1986) was used to assess phonological encoding and syllable sequencing abilities in polysyllabic nonsense words. This task assesses the ability to encode and sequence phonemes and syllables in nonwords that conform to the phonotactic structure of English. Participants were asked to repeat a list of 15

nonsense words after the examiner, and the responses were audiotaped, phonetically transcribed, and analyzed for phonological processes and syllable structure. The percentage of correctly produced words was calculated and transformed into z-scores based on the performance of unaffected siblings of probands (n = 548) assessed as part of a larger longitudinal study, The Cleveland Family Speech and Reading Study. These scores were transformed into standard scores with a mean of 100 and a standard deviation of 15 to be comparable to the other variables in the study and entered into analysis.

Woodcock Reading Mastery Test-Third Edition (WRMT-3) (Woodcock, 2011) was used to assess single-word decoding for both real and nonsense words. The Word Identification and Word Attack subtests comprise the Basic Skills Cluster for the WRMT-3 and provided a broad measure of basic reading skills. The Word Identification subtest requires the participant to read real words of increasing complexity and provides an index of words that are recognizable by sight. The Word Attack subtest requires participants to read nonwords of increasing complexity and measures knowledge of sound-symbol correspondence and application of phonics generalizations. The Basic Skills Cluster scores from the WRMT-3 were used in analysis; they are age-adjusted standard scores with a mean of 100 and a standard deviation of 15. Scores between -1*SD* and +1*SD* are within the average range.

The Test of Word Reading Efficiency-Second Edition (TOWRE-2) (Torgesen, Wagner, & Rashotte, 2012) was used to assess an individual's ability to read both real and nonsense words accurately and fluently under a time constraint. The Sight Word Efficiency (SWE) subtest was used to assess the ability to recognize real words as whole units within 45 seconds. The

Phonemic Decoding Efficiency (PDE) subtest assesses the ability to apply knowledge of phonics and sound-symbol relationships to decode nonsense words within 45 seconds. The test uses age-adjusted standard scores with a mean of 100 and a standard deviation of 15. The Total Word Reading Efficiency Index Score, a combination of SWE and PDE subtests, was used in analysis. Standard scores between 90 and 110 are within the average range.

The Test of Silent Word Reading Fluency-Second Edition (TOSWRF-2) (Mather, Hammill, Allen, & Roberts, 2014) was used to assess the ability to *recognize* printed words accurately and efficiently (i.e., word identification). Speech articulation difficulties may interfere with fluent decoding, and it is important to disentangle the confounding element of speech production while reading out loud. Participants were presented with rows of words, ordered by reading difficulty with no spaces between the words (e.g., dimhowfigblue); they were given 3 minutes to draw a line between the boundaries of as many words as possible (e.g., dim/how/fig/blue). Scores used in analysis were age-adjusted standard scores with a mean of 100 and a standard deviation of 15. Standard scores of 90 to 109 are within the average range. Three participants with CAS taken from the Biomarker of SSD study did not have this assessment; therefore, analysis for this assessment was based on 13 participants with CAS.

The Wechsler Individual Achievement Test-Third Edition, Spelling Subtest (WIAT-3) (Wechsler, 2009) was administered to assess written spelling of letter sounds and single words. Out of the 32 participants, 25 had data for this evaluation. Scores used in analysis were age-adjusted standard scores with a mean of 100 and a standard deviation of 15. Standard scores between 85 and 115 are considered within the average range.

Test of Written Spelling-Fourth Edition (TWS-4) (Larsen, Hammill & Moats, 2011) is a measurement of the written spelling of single words, similar to the WIAT-3 spelling assessment outlined in the previous paragraph. Data from this assessment was used for three CAS participants whose data were collected from the Biomarkers of SSD study. A sensitivity analysis was conducted to ensure that the data for these 3 individuals with different but similar spelling assessments did not influence the results significantly. Scores used in analysis were age-adjusted standard scores with a mean of 100 and a standard deviation of 15. Scores between 90 and 110 are within the average range.

A Test of Children's English and Spanish Speech Perception in Noise (ChEgSS; Calandruccio et al., 2014) was administered to assess masked-speech perception in English only; deficits in speech perception have been cited as contributing factors in both SSD and RD. The ChEgSS is a four-alternative, forced-choice picture-pointing task that can be used in English and Spanish. Target words are disyllabic nouns familiar to young children. The four-alternative format accommodates assessment of children with poor speech production as the tester does not have to assess the child's response. An adaptive track is used to estimate the disyllabic word identification thresholds corresponding to 70.7% correct identification.

All children completed two conditions: (1) English targets in two-talker speech and (2) English targets in a steady-state, speech-shaped noise masker (SSN). The noise was shaped to match the spectral characteristics of the Two Talker masker. The masker stimulus was presented at a fixed level of 60 dB SPL, and the signal level was adjusted based on the listener's responses. The signal level was increased via the MATLAB software after every incorrect response and was

decreased after two consecutive correct responses. These level adjustments were made in steps of 4 dB until two track reversals had been obtained; steps of 2 dB were used thereafter. Each track continued until a total of eight reversals had been obtained, and threshold estimates were based on the average signal level at the last six reversals and the starting masker level to output a threshold signal-to-ratio (dB SNR). Speech reception thresholds were the outcome measures. Lower (better) thresholds indicate that the child can listen at higher levels of noise and still obtain 70.7% correct identification. Three participants from the Biomarkers of SSD study did not have this measure.

Table 2

Experimental Measures Given to All Participants

Domain	Measurement	Purpose	Prediction	Values Reported
Language	PPVT-5	A measure of single-word receptive vocabulary	Vocabulary scores will be depressed in the CAS and RD group	Standard scores
Language	CELF-5 *Core Language Score*	A comprehensive battery of tests that provides a global measure of expressive and receptive language abilities	Language scores will be depressed in the CAS group and RD group	Standard scores
Phonological Processing	CTOPP-2 1) Elision 2) Blending Words 3) Phoneme Isolation 4) Memory for Digits 5) Non-word Repetition 6) Rapid Digit and Letter Naming	A measure of reading-related phonological processing skills	Phonological processing skills will be lower in the CAS group than RD group	Scaled score converted to standard scores

Table Two Continued

Multisyllable Word Repetition	Multisyllable Word Repetition (Catts, 1986)	Measure of lexical retrieval, phonological encoding and accessing motor plans	Scores will be significantly lower in the CAS group	*Z*-score transformed to standard scores
Nonsense Word Repetition	Nonsense Word Repetition (Kamhi & Catts, 1986)	Measure of phonological encoding, memory, and motor planning	Scores will be significantly lower in the CAS group	*Z*-score transformed to standard scores
Decoding Accuracy	WRMT-3 1) Word Identification (WID) 2) Word Attack (WA)	**WID** - An untimed measure of single, sight word reading. **WA** - An untimed, measure of nonsense word decoding	Decoding will be impaired in the CAS group, similar to the RD-no SSD group.	Standard score
Decoding Fluency Oral	TOWRE-2 1) Sight Word Efficiency (SWE) 2) Phonemic Decoding Efficiency (PDE)	**SWE** - Timed measure of single, sight word reading. **PDE** - Timed, measure of nonsense word decoding	Decoding fluency will be impaired in the CAS group, similar to the RD-no SSD group.	Standard score
Decoding Fluency Silent	TOSWRF-2	Measure of ability to recognize printed words accurately and efficiently	CAS group will perform better in silent reading task than oral decoding tasks.	Standard score
Spelling	WIAT-3 Spelling TWS-4	Measure of written spelling of letter sounds and single words.	Spelling skills will be lower in the CAS group.	Standard score
Speech Perception	ChEgSS	Two different masker conditions: Steady-State noise and Two Talker masker	Speech perception will be worse in the CAS group in both SSN and Two Talker masker.	Signal-to-Noise Ratio (SNR) needed for 70.7% correct
Motor-Speech	Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate Total Test	Measure of motor-sequencing skills (rapid repetition of single, bi-syllables and multisyllabic sequences)	Motor speech skills will be lower in the CAS group	*Z*-score transformed to standard scores

3.3.4 Experimental Measures Administered to the CAS Group Only

The Goldman-Fristoe Test of Articulation-Third Edition (GFTA-3) (Goldman & Fristoe, 2015) **Sounds-in-Words subtest** is a measure of an individual's articulation while labeling individual words. It assesses consonants, consonant clusters, and vowel sounds of Standard American English. It was anticipated that some participants with CAS would demonstrate persistent (residual) articulation errors; this assessment was used to compare the speech sound production between the CAS subgroups (Research Question 2). The GFTA-3 uses age-adjusted standard scores with a mean of 100 and a standard deviation of 15. Scores above a standard score of 85 are within the average range.

3.4 Statistical Analyses

3.4.1 Statistical Methods for Research Question 1

Our first research question examined how children with a CAS diagnosis would perform compared to children with RD-no SSD on measures of word-level decoding. To determine the differences between the CAS and RD-no SSD groups on these measures, two-sample *t*-tests (Welch's for unequal variances) or the Wilcoxon rank-sum non-parametric equivalent to the *t*-test were used. The choice of t-test or non-parametric equivalent for the first and second research questions was based on the results of the Shapiro Test, which tests for normally distributed residuals, and the Levene Test, which tests for homogeneity of variances across the groups.

As part of the first research question, we also examined how children with a diagnosis of CAS would differ from children with RD-no SSD on measures of speech sound production, oral language, single word receptive vocabulary, phonological processing abilities, multisyllable

word repetition, diadochokinetic rates, spelling, and speech perception in noise. To determine the differences between the CAS and RD-no SSD groups on these measures, ANCOVA, two-sample *t*-tests (Welch's for unequal variances and Student's *t* for equal variances), or the Wilcoxon rank sum non-parametric equivalent to the *t*-test were used. The choice of *t*-test or non-parametric equivalent was based on the results of the Shapiro Test, which tests for normally distributed residuals, and the Levene Test, which tests for homogeneity of variances across groups. The ANCOVA was used for the speech in noise measure because it allowed for an adjustment in age.

Standardized scores with a mean of 100 and a standard deviation of 15 were used in all analyses, except for the speech-in-noise assessment. For this assessment, speech reception thresholds (dB SNR) at 70.7% correct recognition performance were used for each masker condition. Multiple testing correction was applied separately for Aims 1 and 2 using the methodology of Benjamini & Hochberg (1995). The Benjamini & Hochberg method controls the false discovery rate (FDR)—the expected proportion of false discoveries among the rejected hypotheses. The false discovery rate is a less stringent condition than the family-wise error rate, so it is generally considered more powerful. For each Aim, the total number of independent hypothesis tests were corrected for by using an FDR threshold of .05. For Aim 1, there were 15 independent hypothesis tests. The speech-in-noise domain results included a covariate adjustment for age. Additionally, post-hoc analyses were conducted for the subtests comprising the Phonological Memory Composite. For these subtests a Bonferroni correction for multiple testing was applied.

3.4.2 Statistical Methods for Research Question 2

The second research question investigated if subgroups, defined by below-average decoding fluency on the Reading Efficiency Index from the TOWRE-2 (see section 3.3.3 for a description of this Index on the TOWRE-2) differed on skills associated with decoding ability (i.e., speech sound production, phonological processing skills, and speech perception in noise). To examine this, CAS participants were assigned to groups based on average (i.e., SS $\geq$ 90, CAS-A readers) or below-average decoding performance (SS < 90, CAS-BA readers) on the TOWRE-2. The CAS-A and CAS-BA groups were then compared in the following areas: single-word speech production, PM, PA, and masked speech recognition (in both masker types, SSN and two-talker masker).

To determine the differences within the CAS group on these measures, ANCOVA, two-sample *t*-tests (Welch's for unequal variances and Student's *t* for equal variances) or the Wilcoxon rank sum non-parametric equivalent to the *t*-test were used. The choice of *t*-test or non-parametric equivalent was based on the results of the Shapiro Test which tests for normally distributed residuals, and the Levene Test which tests for homogeneity of variances across the groups. The ANCOVA was used for the speech in noise measure because it allowed for a covariate adjustment in age. Standardized scores with a mean of 100 and a standard deviation of 15 were used in all analyses except for the speech perception in noise assessment. For the speech perception in noise test, speech perception thresholds (dB SNR) were used for each masker condition.

As noted, multiple testing correction was applied separately for Aim 2 utilizing the methodology of Benjamini & Hochberg (1995). For Aim 2, five independent hypothesis tests were corrected for using an FDR threshold of .05.

Chapter 4. Results

4.1 Comparisons between CAS and RD-no SSD Groups (Research Question 1)

4.1.1 Demographic Information

The CAS (n = 16) and RD-no SSD (n = 16) groups were compared on the following demographic variables: age, sex, PIQ, and SES. Two-sample t-tests were used for the quantitative variables, PIQ and age, and Fisher's exact test for categorical variables, sex, and SES. Demographic comparisons between the two groups failed to reveal significant differences in gender (p = .46). There also was not a significant difference between the groups in the mean age of the study participants ($t(27) = -0.31$, p = .76). Non-verbal PIQ did not differ significantly between the groups ($t(27) = -1.32$, p = .20). While SES did differ significantly between the two groups (p = .039), these differences surfaced between the two highest levels of SES and were not considered clinically meaningful (Table 3).

Table 3

Demographic Results Comparing RD-no SSD and CAS Participants

	RD-no SSD (*n* = 16***)***		***CAS*** *(n = 16)*			
Measure (Quantitative)	*Mean*	*SD*	*Mean*	*SD*	*Statistic*	*p*
KBIT, Nonverbal IQ	113.56	12.79	107.06	14.91	-1.32	0.20 [t]
Participant Age	10.99	2.23	10.78	1.59	-0.31	0.76 [t]
Measure (Categorical)	*Freq*		*Freq*		*Statistic*	*p*
Hollingshead SES					NA	0.039 [f*]
1	0		0			
2	0		0			
3	0		2			
4	7		11			
5	9		3			
Sex (% Male)	9 (56%)		12 (75%)		NA	0.46 [f]

Note. Hollingshead ratings: 1 = unskilled laborers, menial service; 2 = machine operators, semiskilled workers; 3 = skilled craftsmen, clerical, sales workers; 4 = medium business, minor professional, technical; 5 = major business and professional.
[t] Student's *t*-test, [f] Fisher's exact test.
[*] $p < .05$

4.1.2 Literacy Domain

The frequency of reading difficulties in the CAS participant group was 63% (10/16). Over half of the participants with CAS demonstrated below-average performance on at least two

of the measures of single-word decoding. This rate indicates a significantly increased rate of RD relative to both population estimates (i.e., 5 to17%) and estimates of RD for children with a history of other idiopathic SSD (i.e., 25 to 30%). Moreover, the analysis failed to reveal significant differences in any of the literacy measures between the groups described as follows and as shown in Table 4 (below).

There was no significant difference between the groups (p = .22) on the Woodcock Reading Mastery Test Basic Skills Composite, a measure of word-level decoding accuracy, with the RD-no SSD group obtaining a mean score in the borderline-average range and the CAS group scoring in the below-average range, and a small effect size (r = .25). The TOWRE-2 Total Word Reading Efficiency Index, a measure of word-level reading fluency, also failed to reveal significant differences between the groups (p = .93), with both the CAS and RD-no SSD groups obtaining mean scores in the below-average range and an insignificant effect size (r = .03). The Test of Silent Word Reading Fluency also failed to reveal significant differences between the groups (p = .99); both groups scored in the below-average range with an insignificant effect size (r = .00). Contrary to predictions, the CAS group did not perform better on the silent decoding fluency measure than on the oral decoding fluency measure; the mean scores on both fluency measures were in the below-average range. Single word spelling skills also did not differ significantly between the groups (p = .66), with both groups' mean scores falling at the lowest end of the average range and a small effect size (r = .11) (Table 4).

Table 4

Literacy Domain Results

Note. [w] Wilcoxon rank-sum test; *ES* = effect size; [+] Sample size for TOSWRF-2 Silent Word Reading Fluency Index was 13 CAS and 16 RD. [#] Sample size for WIAT-3/TWS-4 spelling was 16 CAS and 13 RD. The effect size reported is the Wilcoxon effect size (r): 0.1 – 0.3 (small effect), 0.3 – 0.5 (moderate effect) and ≥ 0.5 (large effect).
[*]Indicates significant after correction for multiple testing (using FDR at 0.05).

	RD-no SSD *(n = 16)*		**CAS** *(n = 16)*					
Measure	*Mean*	*SD*	*Mean*	*SD*	*Statistic*	*p*	*FDR p*	*ES*
TOWRE-2 Total Efficiency Index	83.75	7.92	84.06	19.08	133.00	.87 [w]	.93	.03
TOSWRF-2 Silent Fluency Index[+]	84.88	9.67	88.92	19.81	104.00	.99 [w]	.99	.00
WRMT-3 Basic Skills Composite	86.06	9.45	83.50	17.55	90.00	.16 [w]	.22	.25
WIAT-3/TWS-4 Spelling[#]	88.62	9.42	88.69	19.59	90.50	.57 [w]	.66	.11

4.1.3 Oral Language Domain

The difference between the CAS and RD-no SSD groups' performance on the receptive/expressive oral language measure was not significant. Both groups obtained mean scores within the average range on the CELF-5 Core Language Score ($t(21.72)) = -1.85$, $p = .12$). However, a moderate effect size was obtained for this analysis ($d = .65$). Although nominally significant, after correction for multiple testing, single-word receptive vocabulary also did not differ significantly between the groups ($p = .06$), with both groups scoring within the average

range on the Peabody Picture Vocabulary Test, version 4 or 5 (Table 5). However, a moderate effect size was obtained for this analysis (r = .38).

Table 5

Language and Phonological Processing Domain Results

			RD-no SSD ***(n = 16)***		**CAS** ***(n = 16)***			
Measure	*Mean*	*SD*	*Mean*	*SD*	*Statistic*	*p*	*FDR p*	*ES*
Peabody Picture Vocabulary Test-5	111.00	11.98	101.62	20.43	71.50	.03 [w]	.06	.38
CELF-5 Core Language Score	112.12	11.94	99.50	24.55	-1.85	.08 [wt]	.12	.65
Elision	104.69	13.10	85.62	17.31	-3.51	.001 [t]	.004*	1.24
Word Blending	104.69	10.72	83.12	19.74	35.50	<.001 [w]	.002*	.62
Phonological Memory Comp+	102.88	14.35	85.38	19.63	-2.77	.010 [t]	.021*	1.03
Rapid Symbolic Naming Comp	87.88	11.94	91.62	15.25	0.77	.44 [t]	.55	.27

Note. CELF-5 = Clinical Evaluation of Language Fundamentals-5, ES = effect size; [wt] Welch's *t*-test. [t] Student's *t*-test. [w] Wilcoxon rank-sum test. [+] Sample size for Phonological Memory Composite was 13 CAS and 16 RD-no SSD participants. The effect size reported for measures compared under the Wilcoxon test is the Wilcoxon effect size (r): 0.10 – 0.3 (small effect), 0.30 – 0.5 (moderate effect) and $\geq$ 0.5 (large effect). The effect size reported for measures compared under the student's t-test and Welch's t-test is Cohen's *d* effect size, d = 0.2 is considered a 'small' effect size, 0.5 represents a 'medium' effect size and 0.8 a 'large' effect size.

[*]Indicates significant after correction for multiple testing (using FDR at 0.05).

4.1.4 Phonological Processing Domain

Significant differences surfaced between the two groups in the phonological processing domain. Both subtests measuring PA, Elision, and Word Blending, fell below the normative mean for the CAS group, while the mean for the RD-no SSD group was solidly within the average range —Elision ($t(30) = -3.51$, $p = .004$), Word Blending —($t(30) = -35.50$, $p = .002$). Effect sizes were large for both Elision ($d = 1.24$) and Word Blending ($r = .62$). The Phonological Memory composite was also significantly different between the groups ($t(27) = -2.77$, $p = .02$), with a large effect size ($d = 1.03$), falling in the below-average range for the CAS group and the solidly average range for the RD-no SSD group. In contrast, Rapid Symbolic Naming did not differ significantly between the groups ($t(30) = 0.77$, $p = .55$), with a small effect size ($d = .27$). The mean score for the CAS group fell in the borderline-average range, and the RD-no SSD group fell just below the normative average (Table 5 above).

Post-hoc analyses on the subtests that comprise the Phonological Memory Composite examined potential sources of difficulty within the composite score. As shown in Table 6, after applying a Bonferroni correction for multiple testing, significant differences were found between the groups on the Nonword Repetition subtest ($p = .003$), with the CAS group performing more poorly. Although not statistically significant after Bonferroni correction, a nominally significant difference was found on the Memory for Digits subtest ($p = .04$). The RD-no SSD participants obtained a mean score in the average range, and the CAS group scored below the test's normative average.

Table 6

Phonological Memory Subtest Results

	RD-no SSD (n = 16)			CAS (n = 13)				
Measure	*Mean*	*SD*	*Range*	*Mean*	*SD*	*Range*	*Statistic*	*p*
Memory for Digits[+]	99.69	13.10	[70–110]	86.92	20.16	[55–130]	56.5 [w]	.04
Nonword Repetition[+]	104.38	12.09	[85–125]	88.08	14.37	[55–105]	-3.32 [t]	.003[*]

Note. [wt] Welch's *t*-test. [t] Student's t-test. [w] Wilcoxon rank-sum test.
[*]Significant at the .025 level after a Bonferroni adjustment for multiple testing.

4.1.5 Motor Speech Domain

As expected, there were significant differences ($t(22.32) = -3.89$, $p < .005$) between the two groups on the Fletcher syllable sequencing task, with a large effect size ($d = 1.38$). The RD-no SSD group scored within the solidly average range on the Fletcher time-by-count test of diadochokinetic syllable rate Total Test score. In contrast, the CAS group scored well below the normative average. Similarly, significant differences and a large effect size ($d = 2.08$) surfaced between the groups in repeating multisyllable real words (MSW) ($t(17.87) = -5.88$, $p < .001$), with the CAS group scoring well below the normative average and the RD-no SSD group scoring in the solidly average range. Finally, significant differences ($t(20.71) = -5.43$, $p < .001$) surfaced between the groups when repeating multisyllable nonsense words (NWR), with the CAS group scoring well below the normative average while the RD-no SSD scored in the solidly average range (Table 7). A large effect size was obtained for NWR ($d = 1.92$).

Table 7

Motor-Speech Domain Results

	RD-no SSD *(n = 16)*		**CAS** *(n = 16)*					
Measure	*Mean*	*SD*	*Mean*	*SD*	*Statistic*	*p*	*FDR p*	*ES*
Fletcher (TSS)	101.67	9.78	80.75	19.16	-3.89	<.001 [wt]	<.005[*]	1.38
MSW	111.33	7.90	72.21	25.41	-5.88	<.0001 [wt]	<.001[*]	2.08
NWR	111.29	9.77	78.60	21.99	-5.43	<.0001 [wt]	<.001[*]	1.92

Note. NWR = Nonsense Word Repetition, MSW = Multisyllable Word Repetition, Fletcher = Fletcher time-by-count test of diadochokinetic syllable rate, TSS = Total Test score. ES = effect size. [wt] Welch's *t*-test. The effect size reported is the Cohen's *d* consistent with the Welch's *t*-test. Note that *d* = 0.2 is considered a 'small' effect size, 0.5 represents a 'medium' effect size and 0.8 a 'large' effect size.
[*]Indicates significant after correction for multiple testing (using FDR at 0.05).

4.1.6 Speech-in-Noise Domain

SRTs in the CAS and RD-no SSD groups were compared in two masker conditions, SSN and the Two Talker masker. As shown in Table 8, significant group differences were observed for the SSN SRT (p = .018), with the CAS group performing more poorly than the RD-no SSD group. Additionally, there was a large effect size, supporting meaningful differences between the groups on this measure (eta^2 = .25). As shown in Figure 4, on average, the CAS group demonstrated poorer SRTs than the RD-no SSD in the SSN condition; however, improvement was noted with development.

Significant group differences were not observed for the Two Talker Masker SRT (p = .098); however, the CAS group obtained a lower mean SRT (M = -5.92) than the RD-no SSD

group (M = -9.31), with a moderate effect size (eta^2 = .13). Furthermore, as shown in Figure 5, while both groups demonstrated improvement in SRTs with age, the participants in the RD-no SSD group demonstrated better SRTs than the CAS group with development. Overall, masked-speech recognition performance of the CAS group was poorer than the RD-no SSD group. These findings suggest that when children have a history of deficit in speech production and literacy, they may experience a greater detriment in speech-in-noise perception ability.

Table 8

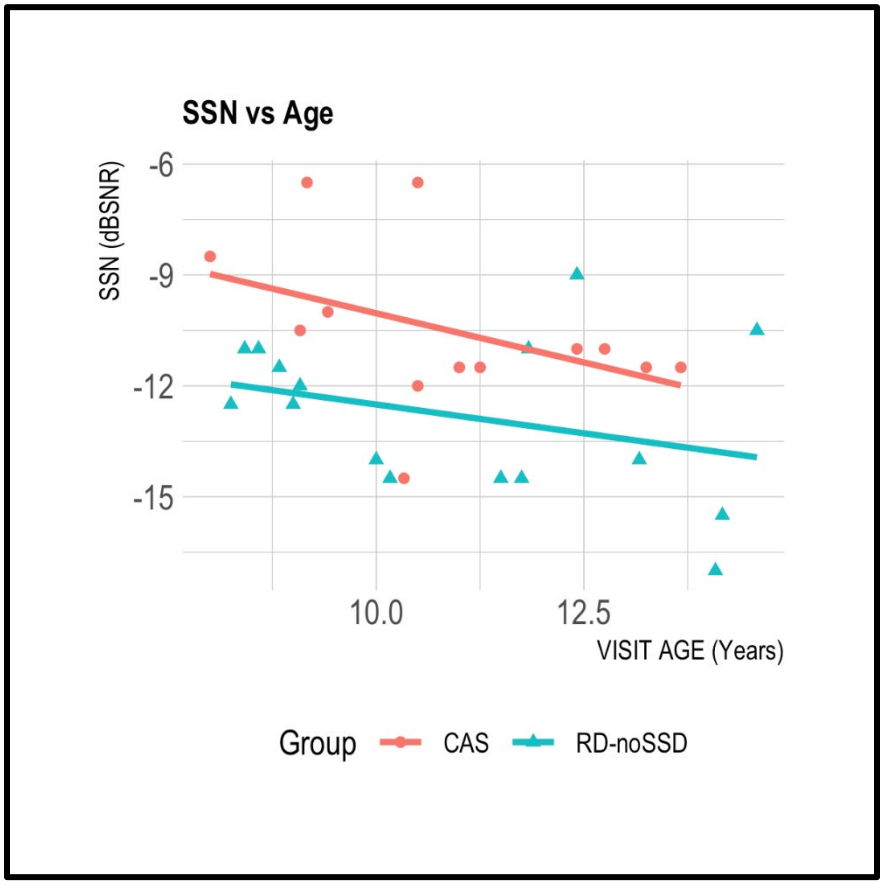
Speech-in-Noise Domain Results

	RD-no SSD *(n = 16)*		**CAS** *(n = 13)*					
Measure	*Mean*	*SD*	*Mean*	*SD*	*Statistic*	*p*	*FDR p*	*ES*
Steady State Noise SRT	-12.81	2.14	-10.50	2.22	8.62	.007 [a]	.018*	.25
Two Talker Masker SRT	-9.31	6.18	-5.92	5.30	3.91	.059 [a]	.098	.13

Note. [a]ANCOVA. Speech in noise domain results include a covariate adjustment for age. ES = effect size. Effect size reported is the generalized Eta-Squared measure of effect size (ges), as reported in ANCOVA. Note that Eta-Squared = 0.01 indicates a small effect; Eta-Squared = 0.06 indicates a medium effect; Eta-Squared = 0.14 indicates a large effect.
*Indicates significant after correction for multiple testing (using FDR at 0.05).

Figure 4

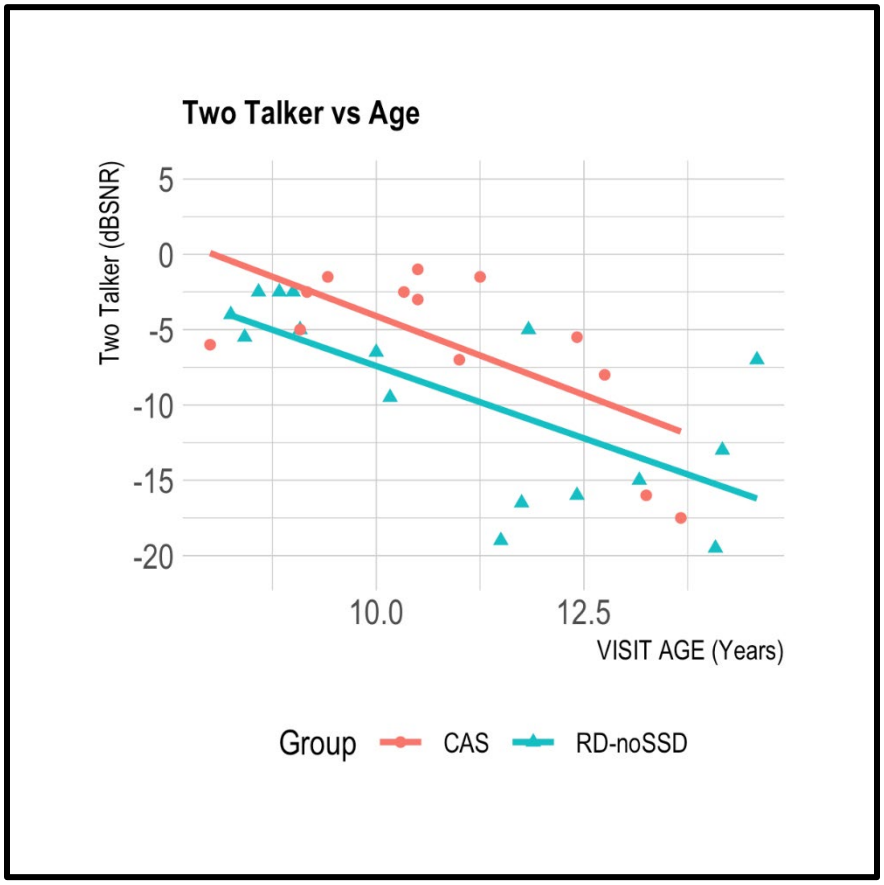
Steady-State Noise SRTs by Diagnostic Group With a Covariate Adjustment for Age



Note. The lines shown in the scatterplot are the least squares regression lines for the association between the speech reception threshold and age.

Figure 5

Two Talker Masker SRTs by Diagnostic Group With a Covariate Adjustment for Age



Note. The lines shown in the scatterplot are the least squares regression lines for the association between the speech reception threshold and age.

4.2 Comparisons Within the CAS Group (Research Question 2)

4.2.1 Demographic Information

The CAS participants were divided into groups based on average ($\geq$ 90, CAS-A, n = 6), versus below-average decoding performance (CAS-BA, n = 10) on the TOWRE Total Word Reading Index Score. As expected, there were significant differences between the groups on this measure (p = .001), with the CAS-BA readers performing worse (M = 74.9) than the CAS-A readers (M = 99.3).

The CAS-A and CAS-BA readers were compared on the following demographic variables: nonverbal PIQ, age, oral language, and SES. Two-sample t-tests were used for quantitative variables of PIQ, age, and language, and Fisher's exact tests were used for the categorical variables, sex, and SES. As shown in Table 9 below, comparisons between the two groups failed to reveal significant differences in non-verbal PIQ (t(14) = 1.38, p = 0.19), age of the study participants (t(14) = 1.19, p = .26), or oral language abilities (i.e., the Core Language measure) (t(14) = 1.75, p = .10), SES (p = .75), and sex (p = .23).

Table 9

Demographic Results Comparing Below-Average and Average Readers With CAS

	CAS-BA Readers *(n* = 10)		***CAS-A Readers*** *(n* = 6)			
Measure (Quantitative)	*Mean*	*SD*	*Mean*	*SD*	*Statistic*	*p*
KBIT, Nonverbal IQ	103.20	15.10	113.50	13.14	1.38	.19 [t]
Participant Age	10.42	1.44	11.38	1.76	1.19	.26 [t]
CELF-5 Core Language Score	91.70	22.51	112.50	23.91	1.75	.10 [t]
TOWRE-2 Total Word Reading Index Score	74.9	12.5	99.3	19.0	60	.001[w*]
Measure (Categorical)						
Hollingshead SES						.75 [f]
1	0		0			
2	0		0			
3	2		0			
4	6		5			
5	2		1			
Sex (% Male)	6 (60%)		6 (100%)			.23 [f]

Note. ratings: 1 = unskilled laborers, menial service workers; 2 = machine operators, semiskilled workers; 3 = skilled craftsmen, clerical, sales workers; 4 = medium business, minor professional, technical; 5 = major business and professional. [t] Student's *t*-test. [f] Fisher's exact test. [w] Wilcoxon Test rank-sum test Hollingshead

[*] $p < .05$

4.2.2 Single-Word Articulation

Statistically significant differences were not found between the CAS-A and CAS-BA reading groups on a measure of single-word articulation (GFTA-3); however, the nominal *p* value was significant, and the corrected *p* value was on the margin of statistical significance ($p = .053$). Furthermore, there was a large effect size ($r = .58$) supporting meaningful differences between the CAS-A and CAS-BA reading groups, with 67% (4/6) of the participants in the CAS-A group demonstrating articulation skills within the normal range, compared to 10% (1/10) of the CAS-BA group (Table 10). Figure 6 shows values for each participant; any score below a standard score of 100 shows persistent articulation errors.

Table 10

Results Comparing Below-Average and Average Readers With CAS

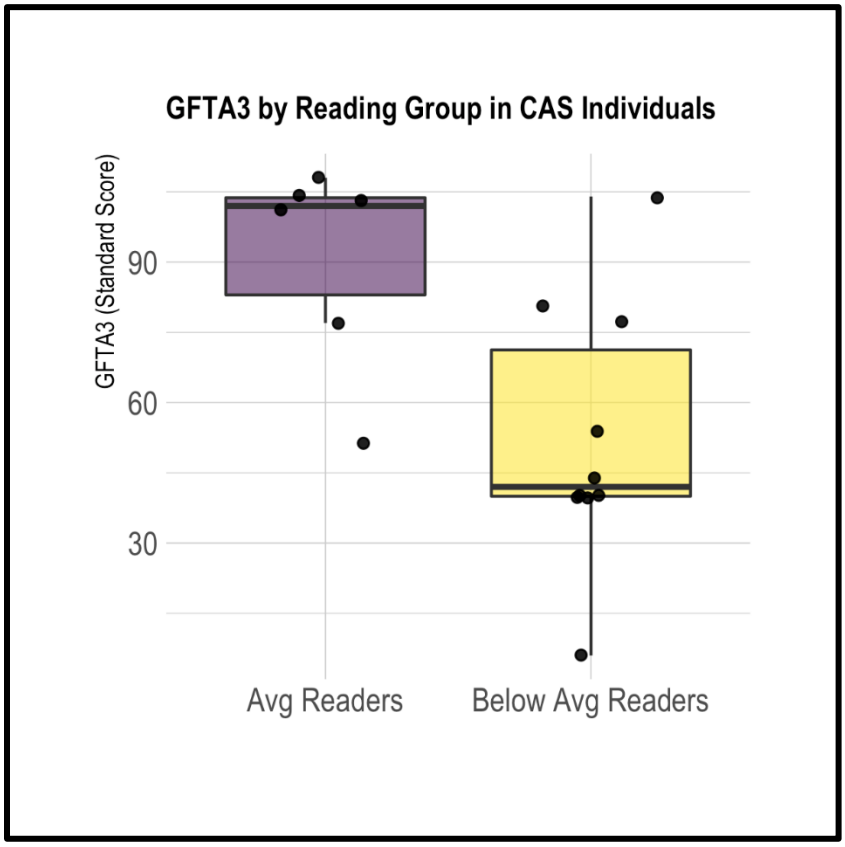
	Below-Average Readers *(n = 10)*		***Average Readers*** *(n = 6)*						
Measure	*Mean*	*SD*	*Mean*	*SD*	*Statistic*	*DF*	*p*	*FDR p*	*ES*
GFTA-3	52.60	27.74	90.67	22.35	51.00	NA	.020 [w]	.053	.58
Elision	78.00	13.58	98.33	16.02	2.72	14	.017 [t]	.053	1.4
Phonological Memory[+]	76.50	17.28	99.60	14.93	2.46	11	.032 [t]	.053	1.4
Two Talker[+]	-3.63	2.56	-9.60	6.740	4.17	1,10	.068 [a]	.085	.29
SSN[+]	-9.63	2.20	-11.90	1.517	2.70	1,10	.13 [a]	.13	.21

Note. GFTA-3 = Goldman Fristoe Test of Articulation-3, SSN = Steady-State noise; Speech-in-noise domain results, SSN and Two Talker, include a covariate adjustment for age. [a] ANCOVA. [t] Student's *t*-test. [w] Wilcoxon rank-sum test;[+] Sample size for Phonological Memory Composite, SSN, and Two Talker masker was 8 below-average readers and 5 average readers. The effect size reported for measures compared under the Wilcoxon test is the Wilcoxon effect size (r). Note that the interpretation values for r: 0.10 – 0.3 (small effect), 0.30 – 0.5 (moderate effect) and $\geq$ 0.5 (large effect). The effect size reported for measures compared under the student's t-test is Cohen's *d* effect size, *d* = 0.2 is considered a 'small' effect size, 0.5 represents a 'medium' effect size and 0.8 a 'large' effect size. Effect size reported for the SSN and Two Talker is the generalized Eta-Squared measure of effect size (ges), as reported in ANCOVA. Eta-Squared = .01 indicates a small effect; Eta-Squared = .06 indicates a medium effect; Eta-Squared = .14 indicates a large effect.

[*]Indicates significant after correction for multiple testing (using FDR at 0.05).

Figure 6

Single Word Articulation Scores by Average and Below-Average Readers With CAS



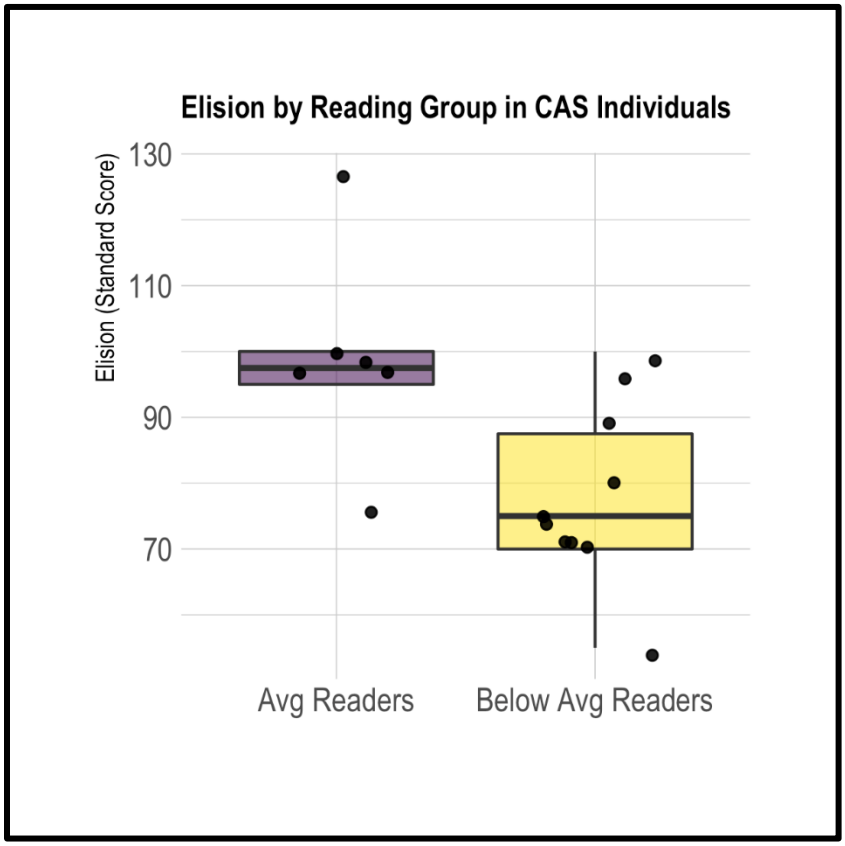
4.2.3 Phonological Processing Skills

As seen in Table 10 (above), a statistically significant difference was not found for Elision (CTOPP-2), a subtest measuring PA ($t(14) = 2.72$, $p = .053$). However, the nominal *p*-value did reach significance, and the corrected *p* value was on the margin of statistical significance. Furthermore, there was a large effect size ($d = 1.4$), supporting meaningful clinical differences. The CAS-A group obtained a mean score within the normative average range ($M =$

98.33), and the CAS-BA group obtained a mean score well below the normative average (M = 78.00). As shown in Figure 7, 17% (1/6) of the CAS-A group fell below the normative average (< 90) on this measure compared to 70% (7/10) of the CAS-BA group.

Figure 7

Phonological Awareness Scores by Average and Below-Average Readers With CAS

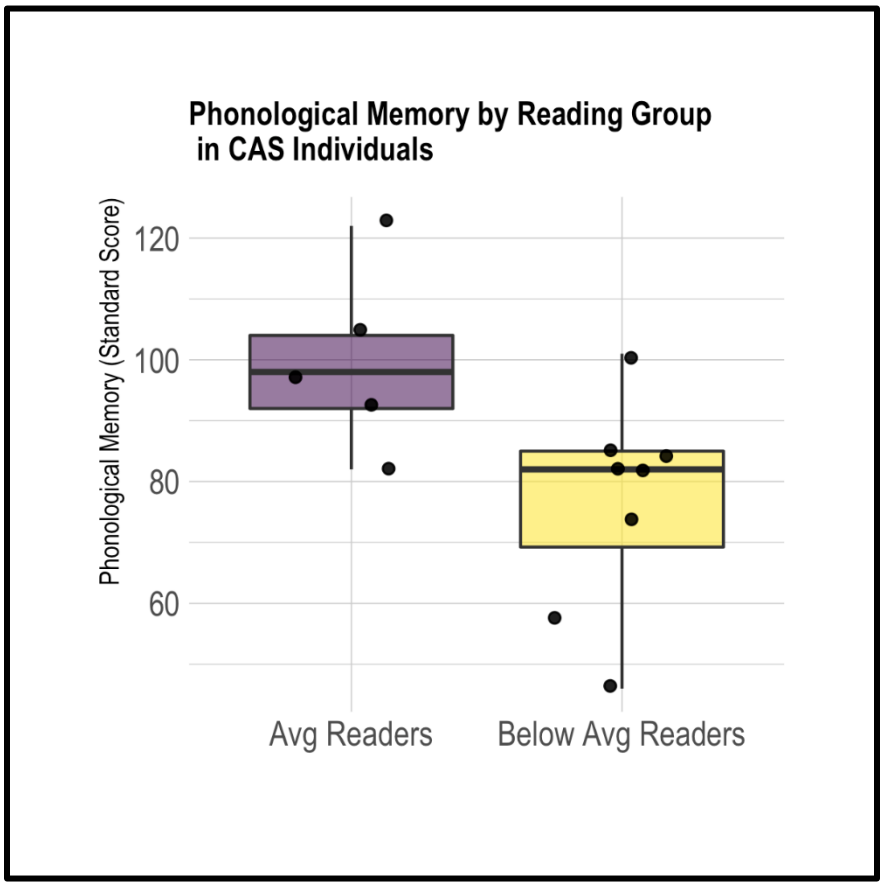


As shown in Table 10 above, PM also did not differ significantly between the groups ($t(11) = 2.46$, $p = .053$); however, the nominal p value did reach significance, and the corrected p value was on the margin of statistical significance. Moreover, there was a large effect size (d = 1.4) supporting meaningful clinical differences between the groups, with the CAS-A group

obtaining a mean score in the solidly average range (M = 99.60) and the CAS-BA group scoring well below the normative mean (M = 76.50). As shown in Figure 8 below, 20% (1/5) of the participants in the CAS-A group obtained scores below the normative average (< 90) on this measure compared to 88% (7/8) of the CAS-BA group.

Figure 8

Phonological Memory Scores by Average and Below-Average Readers With CAS



4.2.4 Speech Perception in Noise

As shown in Table 10 above, the Speech Reception Thresholds (SRTs) were compared in two masker conditions, Steady-State noise, and a Two Talker masker. No significant differences

surfaced between the groups in the Two Talker masker condition ($F(1, 10) = 4.17$, $p = .068$). However, there was a large effect size, indicating possibly meaningful difference between the groups ($eta^2 = .29$). Figure 9 shows the values for each participant. As can be seen in Figure 10, the CAS-A participants' performance appeared to improve with age, which was not the case for the CAS-BA participants. The Steady-State noise masker SRTs also did not reach significance ($F(1, 10) = 2.70$, $p = 0.13$) between the groups; however, once again there was a large effect size ($eta^2 = .21$). Figure 11 shows values for each participant. As can be seen in Figure 12, the CAS-A participants generally had better scores from the outset, while the performance of the CAS-BA participants improved with age.

Figure 9

Two Talker Masker SRTs by Average and Below-Average Readers With CAS

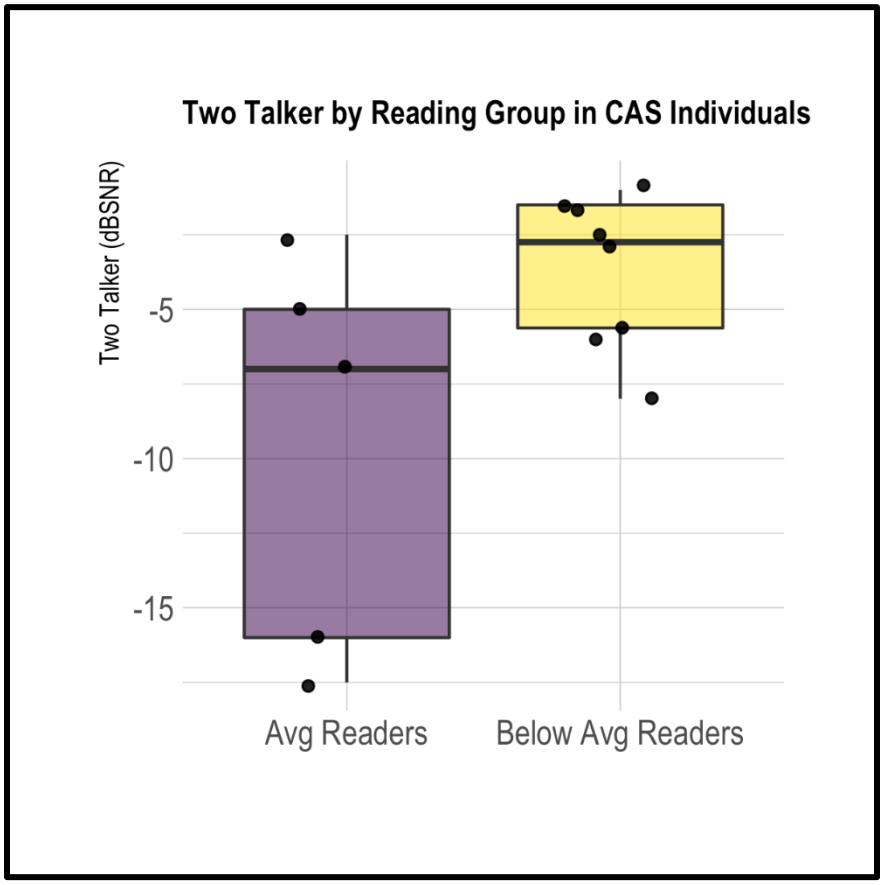
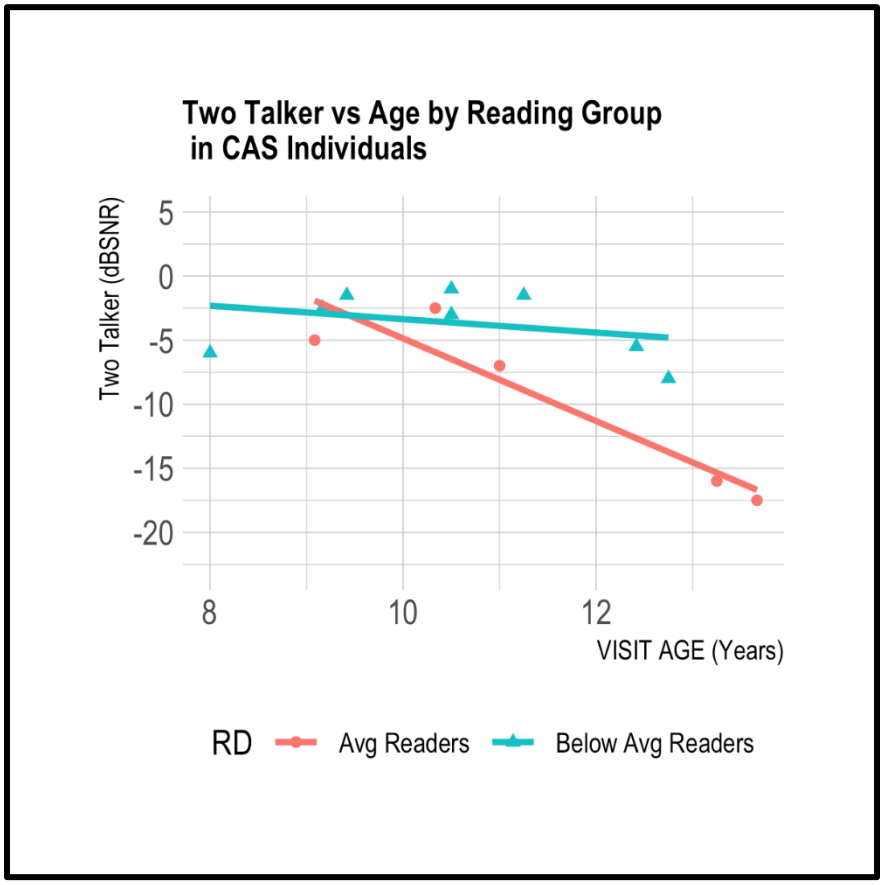


Figure 10

Two Talker Masker SRTs by Reading Group With a Covariate Adjustment by Age



Note. The lines shown in the scatterplot are the least squares regression lines for the association between the speech reception threshold and age.

Figure 11

Steady-State Noise SRTs by Average and Below-Average Readers With CAS

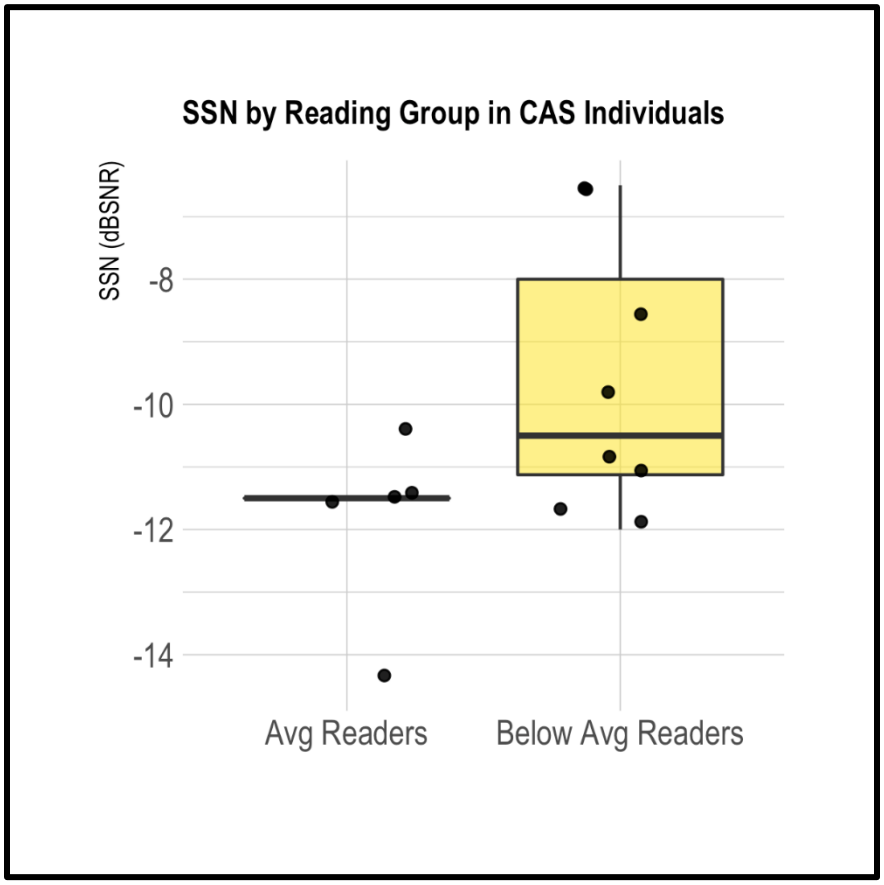
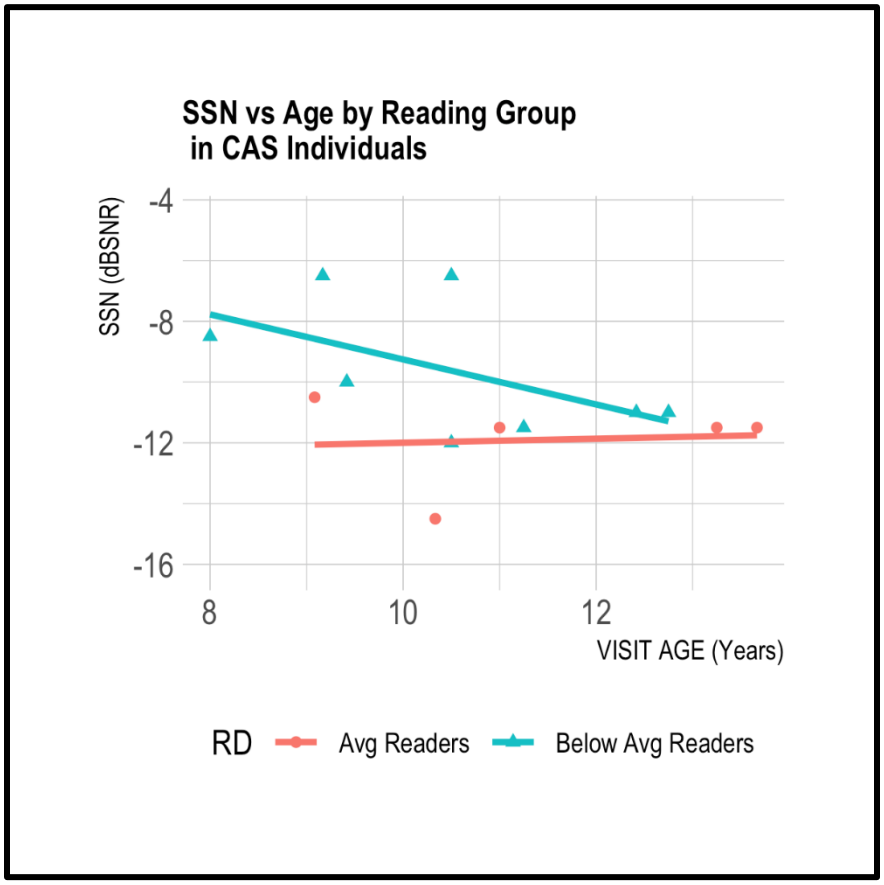


Figure 12

Steady-State Noise SRTs by Reading Group With a Covariate Adjustment by Age



Note. The lines shown in the scatterplot are the least squares regression lines for the association between the speech reception threshold and age.

Chapter 5. Discussion

This section discusses the findings presented in Chapter 4. It is presented in sections corresponding to the study's two aims, divided by the domains examined. Recommendations for clinical intervention follow this discussion.

5.1 Aim 1

The study's first aim was to investigate the frequency of single word decoding impairment for children with a diagnosis of CAS. We hypothesized that children with a CAS diagnosis would demonstrate elevated rates of literacy deficits relative to population estimates and other idiopathic SSD and that participants would demonstrate a similar level of impairment on single word decoding tasks as children with RD-no SSD.

The second part of Aim 1 was to examine whether children with CAS would differ from children with RD-no SSD on the endophenotypes that have been identified to underlie literacy skills. Language skills, including vocabulary, were predicted to be lower in both groups as many children with CAS and children with RD have been shown to have oral language deficits (Catts et al., 1999; Iuzzini-Seigel, 2019; Lewis et al., 2004; McArthur et al., 2000; Wise, 2007). However, language abilities were predicted to be poorer in the group of participants with CAS. We hypothesized that children with CAS would also demonstrate poorer phonological processing skills than children with RD-no SSD. Prior research has implicated a core phonological deficit affecting reading and speech sound production in CAS (Gillon & Moriarty, 2007; Marion et al., 1993; Marquardt et al., 2002; Miller et al., 2019). However, recent research

has not demonstrated that a core phonological deficit is present in *all* cases of RD, especially for those without a prior SSD (Pennington et al., 2012; Peterson et al., 2009; Wolf & Bowers, 1999).

Speech perception in noise was also predicted to be poorer in the CAS group. While some studies have shown a clear relationship speech perception in noise and literacy disorders, (Boets et al., 2008; Ziegler et al., 2009) others have found no relationship between them (Rosen, 2003). On the other hand, children with CAS exhibit difficulties with speech production in noise (Iuzzini-Seigel, 2015), and speech perception has been consistently shown to be a deficit for children with SSD (Hearnshaw et al., 2019; Shiller et al., 2010). Finally, motor-speech skills were predicted to be poorer in children with CAS than children with RD-no SSD; while children with RD demonstrate difficulties with motor-speech sequencing skills (Catt, 1986; Fawcett & Nicholson, 2002), deficits in motor speech sequencing are considered a hallmark of CAS (Peter et al., 2012).

5.1.1 Literacy

The prediction that children with CAS would demonstrate elevated rates of RD relative to population estimates and other idiopathic SSD was borne out in the current study. Sixty-three percent (10/16) of the participants with CAS obtained scores below the normative average on at least two of the three word-level decoding assessments administered as part of the research study. These results are consistent with those reported by Miller et al. (2019), who found that children with CAS demonstrated elevated rates of reading difficulty (i.e., 65%) compared to both population estimates of 5% to 17% (Shaywitz & Shaywitz, 2001), and estimates of RD in other idiopathic SSD of 25% to 30% (Peterson et al., 2009). Furthermore, the present study's findings

also supported the prediction that literacy skills would be similarly depressed in the CAS and RD-no SSD groups. Group means for participants with CAS did not differ from participants with RD-no SSD on word-level decoding accuracy and fluency measures and spelling.

For the measure of word-level decoding accuracy (i.e., Basic Skills Composite, WRMT-3), analysis failed to reveal a statistically significant difference in mean scores between the groups. Sixty-nine percent (11/16) of the participants with CAS scored below the test's normative average on the composite compared to 56% (9/16) of the RD-no SSD participants. To obtain additional information about the reading performance in the groups, we analyzed the results for the subtests comprising the Basic Skills Composite to examine individual participant performance.

Two participants in the CAS group demonstrated a discrepant performance between the subtests (i.e., one subtest score was in the average range and one subtest score in the below-average range). In contrast, five participants from the RD-no SSD group demonstrated a consistent pattern of performance—an average performance on the Word Attack subtest (nonsense words) and below-average performance on the Word Identification subtest (real words). As the majority of the RD-no SSD participants were receiving intervention for reading at the time of the research study, the discrepancy may reflect an emphasis on the remediation of phonemic decoding. Moreover, reduced reading experience (i.e., time spent reading outside of school), often associated with RD, limits growth in the accurate reading of real words (Stanovich, 1988; Torgesen et al., 2012). Guthrie et al. (1999) found that reading amount

predicted reading proficiency, as measured by text comprehension, even when statistically controlling for other variables such as prior topic knowledge and reading motivation.

Word-level decoding *fluency* was measured by the Total Word Reading Efficiency Index of the TOWRE-2, a timed assessment. There was no statistically significant difference in mean scores on this measure, with both groups demonstrating below-average performance. Sixty-three percent (10/16) of the participants with CAS obtained Total Index scores in the below-average range compared to 75% (12/16) of the RD-no SSD participants.

An analysis of performance for the CAS participants on the two subtests that comprise the TOWRE-2 composite revealed that 38% (4/16) demonstrated significantly discrepant scores ($\geq$ 10 points) between the Sight Word Efficiency subtest (SWE, real words) and Phonemic Decoding Efficiency subtest (PDE, nonsense words). Thus, a portion of the CAS participants was better able to decode words that had been previously encountered, consistent with previous research about decoding abilities and CAS (Stackhouse & Snowling, 1992). Regarding the performance of the RD-no SSD participants on these subtests, the timed element of the TOWRE-2 fluency task appeared to remove any benefit of the phonemic decoding strategy that was apparent in the previously described accuracy measure as 81% (13/16) of the participants demonstrated below-average performance on the PDE subtest.

The Test of Silent Word Reading Fluency-2 (TOSWRF-2) is a *silent* measure of fluent word recognition (timed) that mimics real reading. No significant differences surfaced between the CAS and RD-no SSD groups on this assessment, with both groups obtaining mean scores in the below-average range. Of the 16 RD-no SSD participants, 88% (14/16) scored in the below-

average range on this assessment. As predicted, the CAS group performed slightly better on this measure of silent decoding fluency than on the oral decoding fluency assessment; however, of the 13 CAS participants who were administered this assessment, 69% (9/13) obtained scores in the below-average range. Difficulties experienced by the CAS participants on this silent word recognition test demonstrate that motor-speech deficits alone cannot account for issues with decoding.

Single-word spelling skills did not differ significantly between the groups on the Spelling subtest from the Wechsler Individual Achievement Test-3, or the Test of Written Spelling-4. The CAS and the RD-no SSD groups obtained mean scores at the lower end of the average range (M = 88.69 and M = 88.62, respectively). Thirty-eight percent of the participants with CAS (6/16) obtained scores below 1 *SD* of the mean (< 85) compared to 31% (4/13) of the RD-no SSD participants.

Based on the poorer decoding ability in the overall sample, it was expected that more participants would have demonstrated difficulty with spelling. However, there is evidence that single-word dictation, used in the current study, may under-identify children with spelling and written expression difficulties. Mayes et al. (2005) evaluated dictation of single words, sentence composition, and essay composition using standardized assessments in a group of children between the ages of 8 and 13 years (N = 54) who had been referred for a written expression disability. Single-word dictation identified only 28% of the participants as having a written expression disability compared to 78% of the participants who were identified using a standardized essay composition task.

Spelling relies on intact and efficient phonological and orthographic representation and recall (in addition to basic handwriting skills). It involves subvocal rehearsal during which a word is segmented into its component phonemes and converted into graphemes; accurate speech production and intact PA skills are components of phoneme identification. For some of the participants with CAS in the current study, persistent articulation errors and poor PA skills likely undermined spelling. All six of the participants with CAS who obtained scores < 85 on the spelling assessment had significant and persisting errors on the GFTA-3 articulation test.

The present findings are consistent with previous research that demonstrated poorer spelling skills for children with CAS and persisting speech sound errors. Snowling and Stackhouse (1983) examined persistent speech errors, reading, and spelling abilities for four school-age children with CAS (ages 8 to 10;2 years) and compared them to reading-age matched peers. While the experimental tasks appeared relatively straightforward—word repetition, spelling, and reading simple CVC words (e.g., pop, pet, cab)—the children with CAS performed well below the controls on all tasks. Snowling and Stackhouse observed that for the CAS participants during the spelling tasks, "segmentation performance often was faulty and led to the isolation of inappropriate phonemes, prior to phoneme-grapheme translation" (p. 436). Lewis et al. (2004) also found that spelling was particularly impaired in a group of school-age participants with a CAS diagnosis, signaling difficulty in speech sound analysis and segmentation abilities, perhaps due to constraints in PM.

In addition to phonological processing deficits, difficulties with spelling are linked to difficulties with orthographic processing. While reading requires *recognizing* orthographic

elements in a word, spelling involves *recalling* an orthographic representation from memory (Kilpatrick, 2015). Share (1995) proposed a *self-teaching hypothesis* that described phonological decoding as the bootstrap of orthographic representations.

> [E]ach successful decoding encounter with an unfamiliar word provides an opportunity to acquire the word-specific orthographic information that is the foundation of skilled word recognition. A relatively small number of (successful) exposures appear to be sufficient for acquiring orthographic representations, both for adult skilled readers and young children. (p. 155)

Share (1995) suggested that orthographic skill is largely "parasitic upon self-teaching opportunities provided by decoding and print exposure" (p. 169). As most of the participants in both the CAS and RD-no SSD groups demonstrated below-average decoding, it is likely that spelling which requires more detailed orthographic representations than decoding, may remain difficult for some of the participants with CAS and RD-no SSD.

Ehri and Saltmarsh (1995) investigated orthographic representations in a sample of high and low performing first-grade readers and compared them to older disabled third-grade readers by assessing their ability to learn simplified phonetic spellings of words (e.g., messenger spelled MESNGR, stupid spelled STUPD). All participants practiced reading the 16-word list until they mastered the target words. The participants then read the original word list and an altered version (e.g., MESNJR, MEZNGR). The participants' latency in reading the original and then the altered spellings of the practiced words was measured. The younger readers' longer latencies between the original and altered words indicated that they had established orthographic representations of

the original words in memory. The disabled readers did not demonstrate the same latency pattern, suggesting they had acquired only a partial or a degraded orthographic representation of the words in lexical memory. Ehri and Saltmarsh (1995) concluded that deficient word learning processes (i.e., disabled readers took significantly more trials to learn to read the target words) undermine long-term orthographic representations, affecting both decoding and spelling.

5.1.2 Oral Language

5.1.2.1 Language Comprehension and Verbal Expression

The prediction that oral language abilities would be depressed in both groups but lower in the participants with CAS was only partially evidenced in the current study. While the mean score for the CELF-5 Core Language composite, a measure of general language ability, did not differ significantly between the CAS and RD-no SSD groups, the mean scores for both groups *unexpectedly* fell within the average range. However, an examination of individual performance revealed that six of the 16 participants with CAS (38%) obtained scores below 1 *SD* of the mean on this assessment, indicating the likelihood of a language disorder. In contrast, no participants in the RD-no SSD group scored in the disordered range on this assessment. Thus, despite the lack of statistical significance in performance between the groups, a moderate effect size was obtained, indicating meaningful differences in language ability between the RD-no SSD and CAS participants.

Specifically, three of the participants with CAS in the current study demonstrated a global language impairment, with deficits across all four subtests that comprise the Core Language Composite. Three other participants had a mixed profile, with deficits in one subtest in

both the receptive and expressive domains. Research exists that supports the conclusion that receptive language is often superior to expressive language in CAS, and that persistent speech difficulties derail early expressive language skills (Hall et al., 1993; 2006). For example, Hall (1992) stated, "language skills improve after some proficiency with speech sound production" (p. 25). Thus, language difficulties (receptive and expressive) are considered co-occurring rather than an integral part of a CAS diagnosis (ASHA, 2007; Hall, 1992; Hall et al., 1993, 2006; Yoss & Darley, 1974).

Evidence to the contrary also exists. For example, Ekelman and Aram (1983) found that participants, ages 4 through 11 years, diagnosed with CAS demonstrated expressive grammar errors unrelated to speech production skills such as errors in pronoun use, misuse of irregular past tense morphemes, omission of copulas in yes/no questions, and difficulties with question transformations. McNeill and Gillon (2013) investigated receptive and expressive language skills for three children, ages 6;6 through 7;6 years, diagnosed with CAS. Two of the three participants demonstrated a receptive language disorder, and all three had expressive language impairment as shown on the Clinical Evaluation of Language Fundamentals-Fourth Edition (Semel, Wiig, & Secord, 2006). Furthermore, difficulties with morphology and syntax could not be explained entirely by speech errors, implicating linguistic issues as well as motor speech production difficulties in children with CAS (McNeill & Gillon, 2013).

Murray et al. (2019) investigated expressive grammar in preschool children with CAS. Of their 26 participants, 48% had a comorbid morphological disorder. Despite some grammatical errors attributable to speech production, speech difficulties could not explain all the

morphological errors, suggesting that expressive grammar difficulties are co-morbid with CAS and independent of speech production issues. Additionally, even though participants were recruited from previous studies with the requirement of normal receptive language abilities, 19% of the participants demonstrated receptive language impairment. As described earlier, Lewis et al. (2004) assessed a group of 10 school-age children with a diagnosis of CAS using the CELF Language battery; 70% of the participants demonstrated deficits in receptive language, and 80% demonstrated expressive language deficits despite improvement in speech production.

As noted above, no participants in the RD-no SSD group scored in the disordered range on the CELF-5 Core Language Score, indicating that oral language was not impaired for this group of participants with no prior history of an SSD. These findings contrast with McArthur et al. (2000), who reported that approximately 55% of the children diagnosed with specific reading disability exhibited impairments in both reading and oral language. These findings also contrast with Plaza et al. (2002), who found that participants with dyslexia demonstrated significant concurrent impairment in oral language (i.e., word retrieval, syntactic processing, and semantic production) compared to both age-matched and younger-age controls. It is conceivable that the criteria of the current study, i.e., RD-no SSD participants were reported to have no history of speech therapy, reduced the occurrence of oral language deficits in this group of participants. These findings may also suggest that spoken language skills often associated with reading abilities at *younger ages* may not have contributed to the current literacy deficits for the school-age and adolescent participants with RD-no SSD in the present study (Pennington & Bishop, 2009).

5.1.2.2 Receptive Vocabulary

An integral component of oral language ability is vocabulary knowledge, and reductions in vocabulary knowledge are often indicative of a broader language disorder and decoding difficulties (Lyon et al., 2003; Wise et al., 2007). Vocabulary knowledge is also linked to endophenotypes of speech perception, phonological representations, and PA, and is associated with speech production and decoding abilities (Lonigan, 2007; Quinn et al., 2015; Rvachew & Grawberg, 2006). The prediction that receptive vocabulary would be depressed in both groups of participants but lower in the CAS group was not supported in the current study. Mean scores for both groups fell within the average range on either version of the PPVT administered (i.e., PPVT-4 or PPVT-5). However, despite a nonsignificant *p*-value, a moderate effect size was obtained, indicating that meaningful differences surfaced between the groups, with the group of participants with CAS performing more poorly.

There is evidence that single-word receptive vocabulary tests such as the PPVT, often used to screen for issues with lexical acquisition and identify language deficits, do not reliably discriminate between children with normal and impaired language. For example, Gray et al. (1999) investigated the use of tests of single-word vocabulary to differentiate children with a diagnosis of Specific Language Impairment (SLI) and normal controls. The PPVT-3 had a sensitivity rate (accuracy in identifying children in the SLI group as having SLI) of 74% and a specificity rate (accuracy of identifying children in the control group as having normal language) of 71%. The authors proposed that an identification accuracy rate below 80% was unacceptable. While the mean score for the children with SLI in Gray et al. was lower than the controls on the

PPVT-3, their scores usually fell within the "normal" range, calling into question the accuracy of this assessment in identifying deficits in lexical acquisition, similar to the current study. After applying a discriminant analysis function, a cutoff score (SS = 104) for the PPVT-3 was derived that maximized the classification of children into the SLI or control group.

In the current study, 56% (9/16) of the participants with CAS compared to only 13% (2/16) of the RD-no SSD participants obtained a standard score of 97 or below on the PPVT, *well below* the Gray et al. (1999) cutoff score. Three participants in the CAS group obtained scores at or below 1 *SD* of the mean (< 85) on the PPVT; all three participants demonstrated LI on the CELF-5. However, three additional participants with CAS demonstrated LI on the Core Language score but obtained scores within the "expected" range on the PPVT, providing some evidence that the PPVT may not adequately identify language deficits.

Stein et al. (2020) investigated a group of 31 children with CAS using hierarchical cluster analysis for measures of articulation, vocabulary, and reading. Three comorbid subgroups within the CAS participants were identified with varying severity of comorbid difficulties. The highest severity subgroup was characterized by poor reading and the poorest vocabulary knowledge (both receptive and expressive vocabulary). The authors concluded that poor vocabulary knowledge helped identify the children with CAS at highest risk for more severe language and reading disabilities (Stein et al., 2020).

5.1.3 Phonological Processing

Since the 1980s, the most dominant explanation of RD has been a core deficit in the phonological component of language (Lyon et al., 2003; Peterson et al., 2009; Stanovich, 1988).

According to this theory, deficits in *phonological sensitivity* makes learning the correspondences between grapheme and phonemes difficult (Stanovich, 1988). Based on previous research supporting this hypothesis, it was predicted that participants with RD-no SSD would perform poorly on phonological processing measures. It was hypothesized further that participants with CAS would perform even more poorly than the RD-no SSD participants, given research showing the associations between phonological representations, SSD, and PA (Benway et al., 2021; Rvachew & Grawberg, 2006). While there were statistically significant differences and large effect sizes between the two groups on measures of phonological processing, contrary to expectations, the RD-no SSD group obtained mean scores in the solidly average range. Consistent with predictions, the participants with CAS demonstrated mean scores well below the normative average. For the two subtests entered into analysis that are part of the Phonological Awareness Composite (Elision and Word Blending) and the two subtests comprising the Phonological Memory Composite (Memory for Digits, and Nonword Repetition), the RD-no SSD participants obtained mean scores in the average range. In contrast, the CAS participants obtained mean scores below the normative average for all four of these same subtests.

5.1.3.1 Phonological Processing Skills/RD-no SSD

Given the extensive research about the association between deficits in PA and RD, it was unexpected that the participants with RD-no SSD would perform adequately on the PA measures. However, there may be several plausible explanations for these findings. First, the current study's results may differ from previous research because of our population sample. We carefully screened participants with RD-no SSD, documenting an absence of any history of an SSD. In contrast, a review of the extant literature reveals a lack of research in which the variable,

an absence of a history of SSD, has been systematically considered. Because of the difference in chronology between the resolution of an SSD and the emergence of reading difficulty, a child's history of speech sound disorder might not be known to an examiner (Cabbage et al., 2018). Additionally, researchers may consider milder appearing SSD, such as residual articulation errors at reading age, unimportant without an understanding of an earlier, and perhaps, more extensive nature of the speech problem (Cabbage et al., 2018). Thus, there may be differing endophenotypes underlying RD for children with and without a history of SSD that may be blurred when this factor is not considered.

Another plausible explanation for the present findings is that RD may exist as a multideficit disorder without a core phonological deficit in all cases. Pennington et al. (2012) investigated single and multideficit model explanations of dyslexia in two population-based samples. The single deficit models included: (1) a phonological core deficit only, or (2) a single deficit subtype (i.e., a naming speed only, language only, or general processing speed only). The multideficit models included: (1) a phonological core, multiple deficits, multiple predictor model (a single PA deficit is necessary but not sufficient to produce dyslexia, at least two deficits are needed), or (2) a multiple deficit, multiple predictor model (a single deficit is not sufficient to cause dyslexia; at least two deficits are needed; other predictors besides PA would have substantial incremental validity in predicting individual differences in reading skill). The hybrid model predicted subgroups of individuals with dyslexia, some with single deficits and some involving multiple deficits.

Results revealed that only 55% (46/83) of the dyslexic cases had a PA deficit, either alone or in combination with other cognitive deficits in the sample of children from the Colorado Learning Disability Research Center. In the other sample, the International Longitudinal Twin Study, the corresponding rate was 43% (35/82). The authors concluded that "the presence of a PA deficit either to screen for dyslexia or to confirm a dyslexia diagnosis would miss about half the cases of dyslexia" (Pennington et al., 2012, p. 221).

Although our participants were not followed longitudinally, and it is impossible to know whether they had early deficits in PA, another explanation for the average-level PA skills for the RD-no SSD participants may reflect the benefits of remediation. The National Reading Panel (NRP) published its findings in 2000, listing PA as one of the five evidence-based remedial components of reading instruction (National Reading Panel, 2000). Results of the report documented that PA can be taught, and the overall effect size of PA training on outcomes of PA skill was large (0.86). Key elements of the NRP report were implemented into the public schools in the following decade.

For example, the Common Core standard curriculum, adopted by the state of Ohio in 2010 and fully implemented during the 2013–2014 academic year, included PA as a foundational Language Arts skill beginning in kindergarten and continuing into first grade. The PA skills assessed on the CTOPP-2 (i.e., elision, blending words, and isolating phonemes) are currently taught as foundational reading skills. It is very likely that the RD-no SSD participants in the current study, all receiving remediation for RD, also received PA training as part of their regular

curriculum and as part of their remediation programs. Thus, average-level PA skills evidenced by RD-no SSD participants in the current study may reflect the effects of remediation.

Finally, the PA skills assessed by the CTOPP-2 may not capture the level of automaticity required to assess PA's effect on decoding for older children and adolescents with RD. Kilpatrick (2015) suggests that while phoneme manipulation tasks, such as elision, are good predictors of single-word reading, elision can be confounded by a "mental spelling strategy" . . . converting a PA task into a mental spelling task" (Kilpatrick, pp. 158–159). Kilpatrick argues that adding a timed element to phoneme manipulation tasks exposes the accuracy of phonological representations and efficiency of access, revealing important information about a struggling reader's PA. The timing of a participant's response is not a component of the PA tasks on the CTOPP-2.

While there were statistically significant and clinically meaningful differences with large effect sizes between the RD-no SSD and CAS groups for PA and PM skills, there was not a statistically significant difference in mean scores for Rapid Symbolic Naming (i.e., RAN—Rapid Letter Naming and Rapid Digit Naming subtests). The CAS group scored at the lower end of the average range, and the RD-no SSD group obtained a mean score below the standard cutoff score of 90 on the CTOPP-2. Deficits in RAN, the ability to quickly retrieve familiar stimuli in a timed naming task presented in random order and left-to-right serial fashion, have been associated with RD by extensive research (Bowers, 1995; Christo & Davis, 2008; Denckla & Rudel, 1974; Norton & Wolf, 2012).

Additionally, there are differing conceptualizations of how RAN fits into the framework of phonological processing skills. The CTOPP-2 model conceptualizes phonological processing to include PA, PM, and RAN (Torgesen et al., 2012). However, Wolf and Bowers (1999), propose that PA and RAN are distinct constructs and measure different abilities. While RAN includes phonological components such as accessing stored phonological representations and phonological labels when associating them with orthographic information, RAN also includes attentional processes, visual processes (e.g., associating visual features with orthographic representations), accessing semantic information, and speech motor planning leading to articulation (Wolf & Denckla, 2005, Norton & Wolf, 2012; Wolf & Bowers, 1999). Wolf and Bowers (1999) proposed the double deficit hypothesis which separates PA and RAN:

> [P]honological deficits and processes underlying naming-speed deficits represent two separable sources of reading dysfunction . . . Naming-speed and phonological-awareness variables contribute uniquely to different aspects of reading according to this conception, with a model of visual letter naming illustrating both the multicomponential nature of naming speed and why naming speed should not be subsumed under phonological processes. (p. 415)

In the current study, participants with RD-no SSD demonstrated difficulties with the RAN measures, but not PA or PM. Perhaps the PA skills in this group of participants with RD-no SSD improved with remediation; however, fluency-related reading issues are more challenging to remediate. Torgesen et al. (2001) conducted a reading intervention study with third to fifth-grade students. Students made substantial progress in phonemic decoding accuracy,

text reading accuracy, and reading comprehension during the eight-week intervention period; however, their reading fluency showed virtually no improvement. When these same students were followed up two years after the intervention, reading fluency remained well below average, with no improvement despite maintaining gains in the other areas. Torgesen et al. (2001) concluded that lower levels of performance on RAN tasks, compared to PA, at the conclusion of the study were indicative of "fundamental limitations in processing rate for some of the children" (p. 54). Norton and Wolf (2012) describe RAN tasks "as one of the best, perhaps universal, predictors of reading fluency across all known orthographies (p. 430).

One additional aspect of RAN and its relationships to decoding bears discussion— total naming time. Total naming time comprises two components: pause time (the gaps between the articulation of the stimuli) and articulation time (the time taken to articulate the words) (Kirby et al., 2010). Pause time is hypothesized to reflect the automaticity of recognizing stimuli, accessing phonological codes from the lexicon, and shifting attention from one stimulus to the next; articulation time represents the speed in articulating a response once an item has been recognized (Kirby et al., 2010). Al Dahhan et al. (2017) investigated the relationship of single-word decoding, naming speed, and eye movements in three groups of participants: children with dyslexia, ages 9–10; chronological-age controls; and reading-level controls, ages 6–7. Pause time and not articulation time made a significant contribution to reading ability. The authors concluded that RAN is related to poorer single-word decoding fluency by pause time and increased eye fixation durations, reflecting additional time required to encode visual and orthographic information from stimuli.

These findings are congruent with Cutting and Denckla (2001), who found that articulation speed did not contribute significantly to RAN in a group of 79, first through third-grade normal readers. Once a "basal" level of articulation was present, the articulatory demands of RAN are not the critical determinant of RAN speed (Cutting & Denckla, 2001, p. 696). For the participants with RD-no SSD in the current study, it would appear that reductions in RAN were related to pause time.

5.1.3.2 Phonological Processing Skills/CAS

As noted above, the participants with CAS in the current study struggled with PA tasks, obtaining mean scores below the normative average for the two subtests entered into analysis that measured PA on the CTOPP-2. Some research suggests that early motor speech difficulties impact all levels of speech processing in CAS, from phonological representations to the transformation of representations into a motor plan, thus affecting PA skills. However, other research has focused on impoverished phonological representations as the focal point of difficulty in CAS (Marion et al., 1993; Marquardt et al., 2002; Stackhouse & Wells, 1992; Zaretsky et al., 2010). For example, Marion et al. (1993) investigated rhyming skills for four children with CAS and found that the participants demonstrated a severe deficit in rhyming ability across all tasks compared to age, and gender-matched controls with normal language abilities. The authors concluded that CAS involves a prenatal disruption in the neural substrates designed to encode phonemic representations resulting in "an ill-formed phonological representation system" (Marion et al., 1993, p. 148).

Marquardt et al. (2002) investigated syllable perception in a group of six school-aged children (three with CAS and three controls who had normal speech and language and were matched on gender and age). The children with CAS demonstrated difficulty counting syllables and accessing and comparing syllable representations of position and structure. Marquardt and colleagues concluded that CAS is a disorder characterized by an impoverished phonological representation system. Moriarty and Gillon (2006) conducted an intervention study for three children with CAS targeting phonological representations of erred sounds in the context of PA and accompanying speech production activities. Two of the participants made gains in PA that transferred to speech production. Results were considered evidence of the phonological representation theory of CAS since speech production improved with targeted PA intervention.

There is evidence of a reciprocal relationship between PA and PM, as the number of speech sounds that can be held in memory is associated with performance on PA manipulation tasks. For example, Cutting and Denckla (2001) found a significant correlation between memory span and a phoneme deletion task in their study of 79 neurotypical first through third graders, which others have documented (e.g., Brady 1986). Participants with CAS in the current study also demonstrated deficits in PM, with mean scores falling below the CTOPP's normative average for both the Memory for Digits and Nonword Repetition subtests.

Furthermore, intact phonological representations posited to underlie PA are also hypothesized to underlie PM skills. Melby-Lervag et al. (2012) conducted a systematic meta-analytic review of the relationships between phonemic awareness, rime awareness, verbal short-term memory, and children's word-level reading skills. The review included 235 studies with

995 calculated effect sizes, using unselected samples and children with dyslexia. While phonemic awareness was the strongest correlate of individual differences in word reading ability, the authors concluded that the same representations that underlie phonemic awareness also underlie verbal short-term memory tasks— "phonemically structured phonological representations of words in lexical memory are critical determinants of phonemic awareness, immediate memory performance, and learning to read" (p. 341).

However, difficulties with speech production have also been theorized to impact PA and PM for children with SSD and are an essential consideration for children with CAS. PA requires parsing the acoustic stream of a word into separate phonemes. While this usually happens sub-vocally, articulatory gestures are enlisted. Heilman et al. (1996) advanced the motor-articulatory feedback hypothesis of developmental dyslexia, which proposed that awareness—the feeling of correct articulatory gestures—is an essential component of both PA (i.e., segmentation) and PM tasks (i.e., repetition of phonologically complex words).

Therefore, it is not surprising that participants with CAS performed poorly on the Nonword Repetition subtest (PM measure), which requires encoding and sequencing of phonologically novel and complex syllables and the execution of the accompanying motor program for speech production. However, we would anticipate that children with CAS will have less difficulty on the Memory for Digits subtest. This task requires the repetition of single digits that school-age children would be highly familiar with and for which they have well-established phonological representations and motor programs. Poor performance on the Memory for Digits subtest, which was characteristic of the performance for the participants with CAS in the current

study, suggests that some children with CAS may have bona fide deficits in PM in addition to their difficulty with motor-speech production.

Concerning the Rapid Symbolic Naming Composite (i.e., Rapid Letter and Rapid Digit Naming), the participants with CAS scored at the lowest end of the average range. The extant literature reveals limited research about RAN and SSD, particularly for CAS. Tambyraja et al. (2020) investigated the association between a diagnosis of SSD at school-age and risk for reading difficulties using PA, RAN, percent consonants correct in connected speech (PCC), and oral language abilities as predictors. After accounting for language ability, age, and SES, only PA and PCC were significantly associated with an increased likelihood of being identified as at risk for RD (scoring at, or below, 1 *SD* of the mean on a single-word decoding measure). RAN was not a significant predictor of reading risk in this sample of children with SSD. Tambyraja et al. concluded that children with SSD who are at risk for RD may be more appropriately identified as having a phonological impairment primarily.

Burgoyne et al. (2019) examined reading skills in an unselected group of 559 five-year-old children just after school entry and then six months later, using a variety of measures including, oral language, single-word reading, letter-sound knowledge, PA, RAN, and non-verbal IQ. Of the 559 participants, 6.8% demonstrated speech difficulties. After the six-month interval, results demonstrated that speech difficulties were associated with poorer language and reading skills, with a mediated relationship between speech and reading via phoneme awareness skills. There was no evidence that the relationship between speech difficulties and reading was mediated by either letter-sound knowledge or RAN.

5.1.4 Motor-Speech Domain

As predicted, significant differences surfaced between the CAS and RD-no SSD groups on measures that assess motor-speech sequencing abilities with large effect sizes (i.e., syllable sequencing rate) considered a hallmark characteristic of CAS (Shriberg et al., 2012). None of the RD-no SSD participants obtained a score below 1 *SD* of the mean on the Fletcher Time-by-Count Test of Diadochokinetic Syllable Rate compared to 56% (9/16) of the participants with CAS who obtained scores at least 1 *SD* below the mean (< 85). While there is evidence that children with RD exhibit decreased rates of syllable sequencing, the current study did not support this finding. For example, Fawcett and Nicolson (2002) examined the syllable sequencing rates in two groups of dyslexic adolescents (group 1, M^{age} = 13 years; group 2, M^{age} = 16 years). Both groups demonstrated decreased articulation rates and prolonged pauses between repetitions on syllable sequencing tasks relative to the normally achieving controls. The authors proposed that the pauses were indicative of slower access to phonological representations (i.e., motor planning difficulties) and that the decreased articulation rate was related to the slower production of articulatory gestures.

There may be several explanations for the inconsistent findings between the Fawcett and Nicholson study and the current study. First, while their participants were reported to have no overt articulation errors at the time of the study, participants may have had a speech disorder at a previous timepoint, unlike the RD-no SSD participants in the current study. A second possibility for the difference in findings is that Fawcett and Nicholson used SoundEditTM software to analyze the speech stream, allowing for both visual and auditory analysis. The current study

examined tasks using only a digital sound recorder, which may be inherently less accurate. Finally, unlike the present study, which used a total mean score of all nine syllable production tasks, Fawcett and Nicholson measured single syllable and multisyllable tasks separately. Fawcett and Nicholson found that the single syllable rate (i.e., pʌ tʌ, kʌ) effect sizes were larger (slower) than multisyllabic sequences (pʌtəkə). Analyzing separate sequencing rates may have yielded results similar to Fawcett & Nicholson.

Significant differences with large effect sizes were observed between the groups on the multisyllable repetition tasks, MSW and NWR. MSW involves a complex relationship between motor speech ability and cognitive-linguistic processing (Reuterskiöld & Grigos, 2015). As expected, the participants with CAS in the current study demonstrated significant difficulty with both MSW and NWR tasks; sixty-three percent (10/16) obtained standard scores well below 1 *SD* of the mean on both assessments, and an additional two participants scored below 1 *SD* of the mean on either MSW or NWR. As described above, multisyllable repetition tasks measure a complex subset of skills, including phonological encoding, PM, and transcoding (translating a phonological plan into a motor plan), expected areas of difficulty for participants with CAS in the current study.

Previous research has revealed that children with RD also have weaknesses in multisyllable word production (Catts, 1986; Melby-Lervag & Lervag, 2012). As described earlier, Catts (1986) investigated the association of reading ability and multisyllabic word repetition for adolescents with RD and normal controls. The participants with RD made significantly more errors than the controls in producing multisyllabic words. However, in

contrast, none of the 16 participants with RD-no SSD in the present study scored below 1 SD of the mean on either NWR or MSW. Furthermore, 88% (14/16) of the participants had scores on both assessments above a standard score of 100. As Catts did not expressly exclude participants with a history of SSD, as in the current study, participants may have had deficits in phonological processing and motor programming related to earlier SSD, not present for the RD-no SSD participants. Furthermore, the control group used for the current study comprised siblings of probands (i.e., an individual affected with a disorder who is the first subject in a genetic study) who may have had a genetic predisposition for subclinical speech and language issues; therefore, differences between the control group and the disorder groups may have been less apparent.

Melby-Lervag and Lervag (2012) conducted a meta-analysis of studies examining the relationship between dyslexia and nonword repetition. The results showed that children with dyslexia had poorer nonword repetition skills when compared to both chronological-age and reading-level controls; however, the severity of the nonword repetition deficit was predicted by oral language skills. Children with dyslexia and specific language impairment demonstrated the most severe nonword repetition problems. There were no participants in the RD-no SSD group in the current study who showed deficits in oral language. Possibly, differences related to NWR in the present study and previous research are related to the characteristics of the participant sample, such as oral language.

5.1.5 Speech-in-Noise Domain

Prior research has shown that children with CAS demonstrate lower- and higher-order auditory perception impairments affecting speech production (Nijland, 2009; Terband et al.,

2014). We predicted that the group of participants with CAS would perform more poorly than the RD-no SSD participants on the speech perception in noise tasks. Results indicated that in the SSN masking condition, participants with CAS performed significantly worse, with a large effect size, than the RD-no SSD group, requiring a higher SNR to recognize a target word. For the Two Talker masker condition, the participants with CAS performed on average more poorly than the RD-no SSD group, with a medium effect size even though the difference did not reach significance. Overall, results suggest that children with CAS may experience more difficulty than those with RD-no SSD while attending to speech in noisy environments.

The current study is the first study that has investigated the effects of masking noise on speech perception for children with CAS and therefore presents novel findings. Iuzzini-Seigel and colleagues (2015) performed research involving speech production in noise for children with CAS; however, this investigation was related to speech production, not perception. They reported that the speech production of children with CAS was more affected by SSN masking noise than children with other forms of SSD or neurotypical controls. Findings suggested that children with CAS had developed poorer, more vulnerable feedforward programs during early speech development and needed to rely on online auditory feedback to monitor accurate output (Iuzzini-Segel et al., 2015). Masking noise was proposed to interfere with the ability of the participants with CAS to correct their speech production. Terband et al. (2014) suggested that auditory perception impairments in children with CAS play a fundamental role in the emergence of the disorder by interfering with robust systemic mappings required to create accurate and stable feedforward programs for speech production.

Children and adults with RD also show increased difficulty in noise when recognizing speech, a deficit that has been linked to reading by research demonstrating a clear relationship between speech perception in noise, intact phonological representations, PA, and reading ability (Boets et al., 2007; Boets et al., 2008; Dole et al., 2012; Ziegler et al., 2005; Ziegler et al., 2009). However, other data support no association between speech perception in noise and reading difficulties (Brandt & Rosen, 1980; Rosen, 2003). Complicating the interpretation of these data is the suggestion that any association is related to phonological access rather than intact phonological representations (Ramus, 2001; Ramus & Szenkovits, 2008; Hazan et al., 2013). Of interest, none of the studies referenced above provide participant characteristics for speech sound production or information about a participant's history of an SSD. Therefore, a reason for the divergent findings may have resulted from unanticipated participant differences, particularly SSD or a history of SSD.

Finally, there are differences in the developmental trajectory of children's performance in the two disparate masking conditions. SSN is an energetic masker and generates a masking noise that interferes with processing speech signals at the level of the auditory periphery (Corbin et al., 2016). Children's performance in the SSN masker tends to gradually improve until about 10 years of age, with little difference between child and adult performance after that (Corbin et al., 2016). This developmental trend was observed for participants in the current study. A visual inspection of the scatter plot (Figure 4) clearly shows SRTs improving up to 10 years of age, with a general leveling off after that. While the RD-no SSD participants demonstrated lower

(better) SRTs from the outset, a similar trend was observed for the RD-no SSD group, showing that both groups are getting better with development.

In contrast, the Two Talker masker produces informational masking in conjunction with energetic masking; processing involves segregating the target speech signal from background speech and involves selective attention as the listener attempts to separate the two sources (Brady & Calcus, 2021; Brungart, 2001; Corbin et al., 2016). Corbin et al. (2016) reported that SRTs in a Two Talker masker continued to improve until age 13, with what appeared to be a 'break point' in the developmental trajectory between 13 and 14 years of age. At 14 years, the child-adult difference in SRTs disappeared. In the current study, there were six participants between 13;2, and 14;7 years of age (CAS = 2, RD-no SSD = 4). Consistent with previous research (Buss et al., 2020; Corbin et al., 2016), five of the six participants, including both participants with CAS, demonstrated SRTs that were the same or better than the mean SRT (M = -13.1dBSNR) for a sample of 23 adult controls ranging in age from 18;9 to 41;3 years of age (M^{age} = 24.5 years) (Buss et al., 2020).

5.2 Aim 2

The study's second aim was to determine if there were subgroups within the CAS participant group that could be differentiated by reading ability and whether reading ability was associated with a measure of articulation, PA, and speech perception in noise. We hypothesized that children with CAS who were below-average readers (CAS-BA) would perform more poorly than children with CAS who were average readers (CAS-A) in all three areas: articulation, PA, and speech perception in noise.

5.2.1 Reading Subgroups Within CAS Participant Group

The TOWRE-2 Total Word Reading Efficiency Index measures an individual's ability to pronounce real and phonemically regular nonwords accurately and fluently. A standard score of < 90 is used to determine average versus below-average decoding for The Total Index score. Using this cutoff score, two groups emerged, with 63% (10/16) of the participants falling in the CAS-BA group (mean scores on the Total Index, $M = 74.9$) and 37% (6/16) falling in the CAS-A group (mean scores on the Total Index, $M = 99.3$).

The Total Index score comprises two subtests, the Sight Word Efficiency subtest (SWE, i.e., real words) and the Phonemic Decoding Efficiency subtest (PDE, i.e., nonsense words). Both subtests can be stand-alone measures of either sight word or nonsense word decoding. An examination of the SWE and PDE subtest scores for individual participants in the CAS-BA group provided more information about the reading skills of these participants. For eight participants, both subtest scores were in the below-average range. This finding is expected because phonemic decoding and sight word decoding are generally highly correlated; "good phonemic decoding skills are necessary for the growth of a rich sight word vocabulary" (Torgesen et al., 2012, p. 39). For two participants, one subtest score fell in the borderline-average range, and one subtest score in the below-average range. However, the difference in subtest scores did not reach a confidence level of 90% (i.e., > 9 points), signaling an actual statistical difference in scores. Thus, for all the participants in the CAS-BA reading group, the Total Index Score reflected globally depressed decoding skills.

An examination of the subtest scores for individuals in the CAS-A group revealed a different pattern. While two of the six participants demonstrated solidly average scores on both the SWE and PDE subtests, four participants obtained *difference* scores at the 90 to 95% confidence level, which indicated actual differences in phonetic decoding versus sight word recognition. Three participants demonstrated below-average performance on the PDE subtest while scoring in the average range on SWE. Discrepant scores between the SWE subtest and the PDE subtest in favor of SWE indicate a better ability to recognize words that have been frequently encountered. This pattern is consistent with previous research for children with CAS, which found automaticity for decoding familiar words compared to novel words (i.e., if a word is known, it could be correctly decoded or spelled) (Stackhouse & Snowling, 1992, 1992b). The deficits in phonological processing for participants with CAS make it challenging to access grapheme-phoneme correspondences and to "assemble an output phonological code from the individual components" to read novel words (Stackhouse & Snowling, 1992b, p. 291); literacy development is then limited to "lexical knowledge accumulated on a word-by-word basis" (p. 293). Thus, it appears that a diagnosis of CAS is associated with vulnerabilities in decoding regardless of islands of normal performance for sight word recognition.

5.2.2 Speech Production

The prediction that poorer decoding skills would be associated with persistent speech difficulties in the participants with CAS was confirmed in the current study. While the statistical analysis was on the margin of significance ($p = .053$), a large effect size was obtained, suggesting that there were clinically meaningful differences in speech production between the two groups. Ninety percent (9/10) of the CAS-BA readers continued to demonstrate persisting

speech errors, compared to 33% (2/6) of the CAS-A readers. For these participants, scores on the GFTA-3, Sounds in Words subtest ranged from below the 1st percentile to the 6th percentile, indicating significant and persisting speech sound errors for the participant's chronological age. Thus, over half of the participants with CAS (56%, 9/16) demonstrated persisting speech errors and below-average decoding. These findings are consistent with previous research that the persistence of an SSD beyond school entrance increases the risk for reading difficulties (Bird et al., 1995; Bishop & Adams, 1990; Nathan et al., 2004).

A question arises about the contribution of output phonology (articulation difficulties) to reading difficulties (Stackhouse & Snowling, 1992b). In other words, does poor articulation interfere with decoding accuracy? To investigate this, we compared a measure of silent decoding fluency of real words (TOSWRF-2) and the TOWRE-2 subtest of SWE (oral decoding fluency of real words) for the 13 participants with CAS who had undergone both assessments. Seven of eight children who exhibited persistent articulation difficulties demonstrated below-average decoding on the SWE and TOSWRF-2 assessments, demonstrating that poor articulation was not the only source of decoding difficulty.

5.2.3 Phonological Processing (Elision and Phonological Memory)

The current study found that the CAS-BA readers demonstrated greater deficits in phonological processing skills than the CAS-A readers. This finding is not unexpected as a prevailing hypothesis of developmental reading difficulties proposes that poor readers may have heterogeneous cognitive profiles, but there is often a core deficit in phonological processing based on poorly formed or deficient phonological representations. Deficient or impoverished

phonological representations have been posited to be an underlying risk factor in SSD, including CAS (Marion et al., 1993; Marquardt et al., 2002; Velleman, 2011).

The difference in mean scores for the Phonological Memory Composite (CTOPP-2, Wagner et al., 2013) was on the margin of significance (p = .053), with a large effect size, suggesting that despite a lack of statistical significance, clinically meaningful differences surfaced between the two reading groups. The CAS-BA group achieved a mean standard score (*SS* = 76.50), of almost 1.5 SDs below the test's normative average (i.e., a standard score $\geq$ 90), while the CAS-A group's mean standard score was in the average range (*SS* = 99.60). A closer examination of the Phonological Memory Composite is warranted. It is composed of two subtests—Memory for Digits and Nonword Repetition, yielding different types of information about PM.

As described previously, the Memory for Digits subtest requires repeating numbers that school-age children would be highly familiar with and for which they would have well-established phonological representations and motor programs. On the other hand, Nonword Repetition requires encoding of complex phonological information and motor planning to produce complex syllables. Seven of eight participants in the CAS-BA reading group, 88%, obtained scores below the test's normative average (i.e., below a scaled score of 8) on the Memory for Digits subtest compared to none of the participants in the CAS-A reading group, who performed within the test's normative average. Poor performance on the Memory for Digits subtest suggests that some children with CAS may have deficits in PM, in addition to their difficulty with motor-speech production. Notably, all seven participants in the CAS-BA who

scored in the below-average range on the Memory for Digits subtest continued to demonstrate speech sound errors, suggesting that PM is associated with persistent speech errors.

The PA subtest, Elision, revealed the same pattern of difference between the groups as the Phonological Memory Composite, a trend toward significance ($p = .053$), with a large effect size, suggesting meaningful clinical differences. The CAS-BA group obtained a mean standard score (*SS* = 78.00), nearly 1.5 *SD*s below the test's normative average (standard score $\geq$ 90), while the CAS-A reading group obtained a mean standard score within the average range (*SS* = 98.33). Phoneme manipulation tasks, such as elision, are good predictors of single-word reading because they involve accessing, segmenting, deleting, and then synthesizing phonological information into pronounceable words, replicating the process of actual decoding (Kilpatrick, 2015). As noted previously, Melby-Lervag et al. (2012) suggest that phonological representations, i.e., what someone *knows* about a speech sound, underlie both phoneme awareness tasks and verbal short-term memory.

5.2.4 Speech Perception in Noise

The SRTs for the CAS-A and CAS-BA groups were compared in SSN and the Two Talker masker. While there were no statistically significant differences in mean scores between the groups in either condition, large effect sizes were obtained for both masking conditions, suggesting possibly meaningful differences in the groups despite a lack of statistical significance. One explanation for the lack of statistical significance between reading ability and masked speech recognition in the current study is that only two of the CAS-A reading group's participants scored in the average range on both subtests of the composite used to assign

participants. Thus, it is possible that reading proficiency was not different enough between the groups to see significant association. Future research is needed with a larger sample of proficient readers with a diagnosis of CAS to discern significant associations between speech perception in noise and reading proficiency for children with CAS and average and below-average reading proficiency.

While there were no significant differences in mean scores between the groups in either condition, several observations about the participants' performance seem notable. First, the CAS-A group's Two Talker masker SRTs appeared to improve with age, consistent with previous findings regarding the Two Talker masker's developmental trajectory (Figure 10). Conversely, on average, the CAS-BA participants demonstrated a limited maturational effect. A possible explanation for this observation is that the CAS-A group had the two oldest participants, with two individuals in the 13-year age range. These participants obtained SRTs above the mean adult control SRTs (cited previously on page 119). In contrast, the CAS-BA group's two oldest participants did not exceed the 12-year-old age range. It is also worth noting and consistent with previous research that while the CAS-A participants had lower SRT scores from the outset in the SSN condition, the performance of the CAS-BA participants appeared to improve with age (Figure 12).

5.3 Clinical Implications

5.3.1 Participants with CAS

Findings of the current study suggest that an early diagnosis of CAS is often comorbid with literacy impairment at school age. Sixty-three percent (10/16) of the current sample of participants with CAS demonstrated below-average decoding on at least two of the three

composite measures of word-level decoding. If a single assessment was used as a benchmark, 75% (12/16) of the participants with CAS demonstrated below-average decoding. Furthermore, three participants received only two reading assessments (silent reading was not assessed). The frequency of RD might have been higher if all decoding measures had been administered.

Furthermore, there was no significant difference in mean scores between the RD-no SSD group and CAS group on the literacy measures. Mean scores ranged from below average on the single word decoding assessments to the borderline average for the spelling assessment. Thus, literacy difficulties for the participants with CAS did not differ significantly from children diagnosed with RD-no SSD.

In addition to ascertaining a high comorbidity rate of RD for children with CAS, the current study identified endophenotypes that appear to underlie literacy difficulties for these participants. On average, the participants with CAS demonstrated problems with phonological processing skills (i.e., PM and PA) and poorer speech perception in noise abilities than participants with RD-no SSD. Identifying the endophenotypes that underlie literacy deficits for children with CAS has direct implications for assessing and providing intervention services. First, it is essential to evaluate risk factors for RD in this population as early as possible (i.e., preschool age, 3 years and older). It is also important to recognize that a child's reading and spelling difficulties depend upon the level at which the phonological system breaks down (Stackhouse, 1993; Stackhouse & Wells, 1997). A comprehensive evaluation of phonological processing deficits is required to address individual differences. However, this can be a daunting task when assessing a child who may be initially unintelligible and determining the accuracy of

responses becomes a challenge. PA tests that do not require a verbal response, such as The Phonological Awareness Test (Bird et al., 1995), might be helpful when evaluating PA skills for children with unintelligible speech.

Evaluating PM poses similar challenges for children with intelligibility constraints. For example, nonword repetition is frequently used to assess PM; however, children with CAS have motor sequencing and speech production difficulties which can confound results. The Syllable Repetition Task (SRT, Shriberg, et al., 2009, www.waisman.wisc.edu/phonolgy/) was developed specifically for children with speech production difficulties. It uses a single vowel and a limited set of early developing consonants in the multisyllable repetition stimuli. The Memory for Digits subtest (CTOPP-2, Wagner et al., 2013), used in the current study, may also help circumvent speech intelligibility issues. It requires repeating single digits that many young children are familiar with and for which they may have well-established motor programs

Similar to current recommendations for children with dyslexia, early intervention is key in lessening the effects of RD for children with CAS. Systematic PA training has been shown to enhance *phonological sensitivity* and *phonological storage mechanisms* underlying phonological representations (van Kleeck et al., 2006), positively impacting literacy development and oral language (Zaretsky et al., 2010). McNeill et al. (2009a) propose an integrated phonological approach that combines targeted speech production practice and training in PA with sound-symbol associations to improve decoding skills for school-age children with CAS. The integrative phonological approach has also been used successfully within a preventative framework for preschool children with a diagnosis of CAS (McNeill et al., 2009). Activities

included targeted phoneme identification and speech production with sound-symbol correspondence. Moderate treatment and large generalization effects for speech production were noted using the integrated phonological approach (Murray et al., 2014), with improvements reported in PA and nonword decoding (McNeill et al., 2009a).

The effect of masking noise on speech perception in school-age children and adolescents with a CAS diagnosis was a novel finding of the current study. This is the first study of speech perception in noise for children with CAS (cf. Iuzzini-Seigel et al., 2015, who investigated speech *production* in noise) Speech perception in noise was poorer in the Two Talker masking condition, on average, and significantly poorer in the SSN masker for participants with CAS compared to the RD-no SSD group. These findings are noteworthy as there is a wealth of research linking deficits in speech perception and production for children with SSD (Shiller et al., 2010; Hearnshaw et al., 2019), and research linking deficits in speech perception, PA and literacy skills for children with SSD (Rvachew, 2007; Rvachew & Grawberg, 2006). Furthermore, there is research that links speech perception in noise to both PA and reading ability (Ziegler et al., 2009). Vanvooren et al. (2017) conducted a longitudinal study of 5-year-old children at high and low familial risk for dyslexia, using speech perception in noise measures. Kindergarten performance in the SSN masker was the largest predictor of phonological awareness at the start of first grade, explaining 12.5% of the variance. Additionally, apart from family risk, it was a unique predictor for reading, explaining 6.5% of the variance.

Given the association between deficits in speech perception, production, and PA, literacy remediation that focuses on speech perception might improve reading-related skills and reading

ability of children with CAS. For example, Gonzalez et al. (2002) demonstrated significant improvement in reading skills for a group of 9- to 11-year-old children diagnosed with RD compared to controls (children with RD who did not receive the experimental interventions). One treatment group received an intervention that included speech discrimination, letter-sound correspondence, and phonemic awareness training, compared to intervention consisting of letter-sound correspondence and phonemic awareness. Only the intervention group that received speech perception training improved their reading skills significantly compared to the controls.

Additionally, perceiving speech in noisy environments is a developmental skill (Bradley & Sato, 2008; Corbin et al., 2016). The youngest children, whose classrooms tend to be noisiest, are the most susceptible to real-life classroom noise (Jamieson et al., 2004). Until these skills are fully developed, young children expend cognitive effort parsing a speaker's message in a noisy classroom, thus depleting their available resources for learning, including vulnerable prereaders, and beginning readers (Bradley & Sato, 2008; Sato & Bradley, 2008; Shield & Dockrell, 2008). Classroom background noise levels often exceed the American National Standards Institute/Acoustical Society of America's (2010) Standard S.12.60 recommendations. These levels are especially deleterious to children with normal hearing and learning difficulties, such as speech and language delays and reading disorders. Recommendations to reduce noise levels in classrooms include reducing ventilation noise and other noise sources, applying construction specifications to control sound transmission and reverberation, and increasing the availability of sound-field FM amplification systems, thereby improving the SNR throughout the classroom environment for the benefit of all students (Jamieson et al., 2004).

Finally, the persistence of speech sound errors was associated with deficits in decoding skills for the participants with CAS. Eighty-two percent (9/11) of the participants with persistent speech errors demonstrated RD. Thus, an early diagnosis of CAS in conjunction with persisting speech sound errors could be considered a strong indication that a child is at risk for reading difficulties. However, while five of the participants with CAS in the current study demonstrated normal speech sound production, only two of these participants demonstrated average single word decoding across *all* of the decoding assessments. These findings have direct implications for monitoring and intervention for special education services. First, many children with CAS continue to be enrolled in speech-language therapy at school age due to persistent speech and language problems. In addition to providing intervention for speech sound production, language, and preliteracy skills, speech-language clinicians need to carefully monitor progress in PA and decoding skills to ensure a timely referral for a literacy assessment if necessary. Furthermore, it may be unwise to remove children with a diagnosis of CAS from an IEP, even if their speech normalizes, when there is evidence of phonological processing or decoding difficulties. These same children are at risk of being placed back on an IEP by middle school because of learning issues, especially reading.

5.3.2 Participants with RD-no SSD

The current study revealed that all the RD-no SSD participants demonstrated phonological processing skills (i.e., Phonological Awareness Composite) within the CTOPP-2's normative average range, except for RAN. Fifty-six percent (9/16) of the RD-no SSD participants scored below the test's normative average, rapidly retrieving both letters and numbers. As described earlier, lower performance levels on RAN tasks, compared to PA,

indicate underlying limitations in processing rate associated with reading fluency (Torgesen et al., 2001). Norton and Wolf (2012) describe RAN tasks as universal predictors of reading fluency, measuring shared naming, and reading processes. Additionally, children appear to be "relatively consistent in their overall naming ability across time, relative to peers" (Norton & Wolf, 2012, p. 436). Therefore, improving reading fluency is a complex and time-intensive process.

Morris et al. (2011) investigated the use of multicomponent reading intervention to improve fluency in a group of 279 children diagnosed with a reading impairment, varying by IQ, race, and socioeconomic status. Groups were randomly assigned to one of four interventions, two multicomponent and two contrast interventions. The multicomponent interventions provided training in PA, vocabulary knowledge, morphology, and strategy use (i.e., sounding out words, peeling off affixes, and trying different vowel pronunciations). The contrastive interventions targeted PA and curriculum training or PA and math training. The study revealed that multicomponent interventions were associated with better reading fluency and reading comprehension than the contrastive interventions, regardless of IQ, SES, or race. Previously, these skills have proven challenging to remediate.

In addition to addressing a multicomponent of skills for effective reading fluency intervention, intervention needs to be sufficiently systematic and intensive to be successful. Children in the Morris et al. (2011) study received an hour of instruction per day over 70 days. According to Al Otaiba et al. (2018), children with RD require daily instruction and “frequent opportunities to respond and to receive corrective feedback" (p. 833). The Taxonomy of

Intervention Intensity proposed by Fuchs et al. (2017) outlines seven principles of measuring how effective an intervention might be for a given student. One of the essential principles of the Taxonomy, the dosage dimension, is based on the number of opportunities a student has to respond and receive corrective feedback. The dosage dimension is formulated on the size of the instructional group, the number of minutes each session lasts, and the number of sessions provided per week.

For children with a diagnosis of CAS and comorbid RD, and all children with a diagnosis of RD, reading intervention needs to be sufficiently frequent and intensive, and a student's response to the intervention needs careful monitoring to ensure that their individual needs are being met. Vaughn et al. (2009) recommend that reading intervention be delivered individually or in small groups for extended daily sessions over an extended period for younger students at the highest risk of reading failure. Sessions need to include explicit, systematic word-level instruction, high levels of active student engagement, and practice reading-connected text. Effective intervention for older students with more severe reading difficulties requires even higher doses of intensive intervention over several years (Vaughn et al., 2009).

5.4 Limitations and Future Directions

Several limitations of the current study should be noted. First, the sample size for both the CAS and RD-no SSD groups was small and may have been underpowered to detect differences between the two reading groups. A larger sample size for the CAS group would have allowed for increased power to confidently identify subgroups and detect differences between the CAS-A and CAS-BA participants. For example, the small sample size may have reduced the

representation of individuals with CAS with average reading proficiency, leading to a Type 1 error. Similarly, the small sample size for the RD-no SSD group may have underrepresented individuals with greater PA and PM deficits.

Since this study was not longitudinal, CAS was determined by historical report rather than direct testing; therefore, confirmation of a CAS diagnosis may have been less reliable. Furthermore, the extent of early PA skills was unknown for the RD-no SSD participants, precluding comparisons between possible early difficulties and current performance. Although all participants received speech therapy or reading intervention, as reported by parents, information was not available on the quality and content of intervention received by study participants.

We did not assess reading comprehension or control for other diagnoses such as ADHD. Future research might consider the addition of other comparison groups such as RD+SSD, SSD-only, SSD+LI, and LI-only as it would help to determine if endophenotypes that impact reading in CAS are similar to those in other speech and language disorders. Although all the assessments used in the current study, with the exception of speech perception in noise, were standardized and norm referenced, the inclusion of a control group of typically developing children would have also been a useful comparison, especially for the speech perception in noise measures.

Speech output tasks were used to measure PA skills in the current study. Future studies might examine PA skills for children with CAS by including measures that do not require speech production. The Lindamood Auditory Conceptualization Test (LAC-3; Lindamood & Lindamood, 2005) assesses the ability to manipulate syllables and phonemes in nonwords

without any speech output by having a child manipulate colored blocks and colored felt squares. Similarly, it can be challenging to separate a child's performance on measures of PM from speech production deficits. Along these same lines, it may be helpful to extend the use of silent, word-level decoding measures to include silent sentence-level decoding measures for children with CAS to eliminate the confounding effect of speech production difficulties on reading aloud.

Finally, further research is required to examine the extent of an auditory perception impairment for children with CAS using speech-in-noise tasks. As ChEgSS uses a closed set of words that the participant is familiarized with before assessment and does not require a spoken response, it is a valuable tool in assessing speech perception in noise abilities for children with speech sound production difficulties. Future research might compare the current study's findings (i.e., children with CAS and children with RD-no SSD) to children without any history of reading, language, or speech production difficulties (age-matched controls), using the ChEgSS assessment. This approach will help establish normative data for speech perception in noise abilities for these two disorders. Additionally, collecting data for older participants with CAS and RD-no SSD (i.e., beyond 13 years) might help ascertain whether individuals with these disorders show a similar abrupt improvement in speech perception in noise abilities in the Two Talker masker that has been previously demonstrated.

5.5 Conclusion

Childhood Apraxia of Speech is a developmental speech sound disorder notable for its severity and persistence of speech difficulties. It has been viewed historically as a motor speech disorder, which has limited investigation into literacy skills associated with the disorder.

Nonetheless, children with CAS often present with comorbid language problems (receptive and expressive), learning disabilities (e.g., reading and spelling), and fine and gross motor difficulties. While extensive research has been conducted about the association between a diagnosis of other idiopathic SSD and literacy outcomes, demonstrating a significantly increased risk of reading difficulties, there is limited research investigating this same association for CAS.

This dissertation research sought to bridge this gap by examining literacy skills (i.e., decoding and spelling) and the speech-language correlates of literacy (i.e., phonological processing skills, oral language, speech sound production, multisyllable sequencing and word repetition, and speech perception in noise) in a group of participants with CAS. We compared these participants to children diagnosed with reading disorders but with no history of SSD (RD-no SSD) to determine if these groups differ in literacy skills and ascertain what an SSD might contribute to literacy difficulties by comparing the underlying skills (endophenotypes) associated with literacy. Results suggest that many children with CAS share the same degree of difficulty with word-level decoding and spelling as those diagnosed with reading difficulties who have no history of speech problems. In contrast, phonological processing abilities, crucial for developing reading skills, were significantly reduced for the participants with CAS. These findings suggest that deficits in the underlying skills needed for literacy may differ for children with histories of CAS and require interventions tailored to these specific skills when addressing their reading difficulties.

Furthermore, subgroups within the CAS participants were differentiated by average (CAS-A) and below-average (CAS-BA) word-level decoding fluency. The CAS-BA group

presented persistent speech sound errors and more pervasive PA and PM deficits. Finally, children diagnosed with CAS whose speech does not normalize by school-age appear at heightened risk for literacy difficulties. However, speech normalization does not ensure that literacy skills will develop normally; weaknesses in real and nonword decoding were noted even for those whose speech had normalized.

Appendix A - Informed Consent

IRB NUMBER: STUDY20180545
IRB APPROVAL DATE: 1/11/2019

CASE WESTERN RESERVE UNIVERSITY EST. 1826

INFORMED CONSENT DOCUMENT

Speech and Reading Study - SpARS

Introduction/Purpose

Your child has been invited to participate in a study of speech, language, and reading disorders because he or she has a history of a speech, language or reading disorder. This study will look at behavioral factors that contribute to a reading disorder. The present study is also designed to search for the gene or genes that are responsible for speech, language or reading disorders. There is evidence that speech, language and reading disorders are genetic (inherited or “run in families”) and may be caused by several genes and environmental influences. The genetic studies will provide a "clue" as to the possible location of a gene.

Taking part in this research is completely voluntary. It is important that you read and understand the following information about the study before you decide whether to have your child participate or not. This informed consent form describes the purpose, procedures, benefits, risks, discomforts, and precautions of the study. It alsodescribes the alternative procedures that are available to you and your right to withdraw from the study at any time. No guarantees or assurances can be made as to the results of this study. Please take time to make your decision and discuss it with your doctor, family and friends if you wish.

Your child will be one of 40 participants in this study that is being conducted at Case Western Reserve University.

Participating in research is voluntary, which means the choice is up to you. Choosing not to have your child participate will have no effect on the medical care that they receive. This form, called a consent form, explains what will happen to your child if you decide to allow him/her to participate. Please read it carefully and have all of your questions answered before you make your decision. If you decide to participate, we will have you sign this consent form. You will be given a copy of this form to keep.

Study Procedures

If you agree to participate, your child will be given a speech, hearing and reading screening; and, more extensive speech, language reading, cognitive, and speech in noise tests some of which are similar to those encountered in school. Your child will be audio recorded as part of the speech and language tests. In addition, one or both parents will be asked to complete a questionnaire about your child’s speech, language, and reading disorders. It is possible that your child may not qualify for this study if he/she has any developmental or medical conditions that preclude his/her participation. Your child’s ineligibility may not be discovered until after they complete the screening process. Your child will be compensated for any portion of the evaluation that they complete.

If you agree to have your child participate they will be asked to do the following things:

a) Your child will be asked to listen while a researcher explains an informed assent form and then sign it.

b) You will be asked to fill out a developmental questionnaire about your child's developmental history.

c) Your child will undergo a hearing screening where they will be asked to raise their hand in response to hearing very soft tones (or beeps) at different frequencies (the equivalent of a clinical hearing test). The sounds will be presented over headphones. This is not a medical exam, and is only being conducted for research purposes to confirm that your child has normal hearing on the day of testing. The Principal Investigator will notify you if we find that your child has difficulty hearing some of the tones and provide you with an appropriate recommendation for follow-up.

d) Your child will undergo a 1-hour hearing, speech and/or reading screening which will involve listening to sounds, naming pictures, repeating words or reading single words.

e) If your child passes the hearing, speech and/or reading screening they will undergo a more extensive speech-language, cognitive, reading, and speech-in-noise assessment where they will be asked to name more pictures, listen to words and repeat them, point to pictures, follow directions and read words.

f) During the speech-in noise testing, your child will be asked to listen to words and point to pictures. The sounds your child will be listening to may be presented in quiet or in combination with background sounds such as people talking. The sounds will be presented over headphones at a comfortable listening level (similar to the level of typical conversation).

g) You will receive a report with your child's speech, language, and reading scores.

h) This study will take place during one day of testing at Case Western Reserve University (a 1-hour screening followed by a 3.5-hour testing session for speech-language and reading tasks). The 3.5-hour testing session can be completed with or without a lunch break. You will be given a $25 gift card for restaurants that surround the Cleveland Hearing and Speech Clinic if you choose to take a lunch break.

i) Your child will also be asked to provide a DNA sample through a saliva (spit) sample of about 2 milliliters (about half a teaspoon). He/she will be asked not to eat and to brush their teeth at least 30 minutes prior to the saliva collection. Your child will spit into a small cup. DNA will be obtained from your child's saliva sample and used for the study described. The DNA samples will be stored and analyzed in the laboratory of Dr. Sudha Iyengar here at Case Western Reserve University. Samples collected will be stored indefinitely for future use. This is an optional part of the study and you can still participate in the entire study even if you choose not to have your child provide a DNA sample. If your child's participation is limited to the screening, you will not be asked to provide a DNA sample.

You (or your child) can choose to stop participating for any reason at any time.

Risks

The risks and discomforts associated with this study are limited and do not involve any physical risk to your child except the normal anxiety and fatigue of testing.

There is a risk of breach of confidentiality which means that someone who is not listed in this form might view your data either by accident or from malicious actions they take to hack the data. We are protecting against this by only storing information that can be directly linked to you on CWRU secure computers, in password protected files which are behind firewalls. Measures to protect against loss of confidentiality are described below.

Benefits

You will receive a free speech-language and reading evaluation which may assist in the planning and intervention in your child's academic program. Your child's participation in this study may lead to information regarding behavioral factors related to reading disorder and the specific hereditary nature of speech, language and reading disorders. This information may benefit future generations of families with speech, language and/or reading disorders by providing early and accurate diagnosis.

Alternatives to Study Participation

Because of the nature of this research the only alternative is to not participate in this study. You are free to withdraw or refuse to participate in this study at anytime.

Financial Information

There is no cost to you or your insurance for participation in this study. Your child will receive fifty dollars for your child's participation in the behavioral testing and fifty dollars for your child's participation in the DNA collection. It will be paid in cash. If your child completes all of the behavioral testing and they wish to take a break for lunch midway in the testing, they will receive a $25 gift card to be used for lunch at local restaurants close to the CWRU testing location. There is no reimbursement for gas mileage. If you withdraw from the study, your child will be paid for the portions that he/she has completed. For example:

1) If your child completes the screening but is ineligible for the study, your child will receive $50.

2) If your child completes all of the behavioral testing, they will receive $50 dollars.

3) If your child completes the DNA portion, they will receive an additional $50 dollars.

4) If your child wishes to take a lunch break midway through the behavioral testing, they will receive a $25-dollar gift card for lunch at nearby restaurants.

Use of Samples Data in the future:

DNA will be obtained from your child's saliva sample and used for the study described. The DNA samples will be stored and analyzed in the laboratory of Dr. Sudha Iyengar here at Case Western Reserve University. Samples collected will be stored indefinitely for future use. You have the right to withdraw from this study. If you choose to withdraw no further studies will be completed on the samples that you have contributed. You may withdraw your DNA sample from the study by contacting Dr. Barbara Lewis at (216) 368-4674, or Gabrielle Miller at 781-640-1044.

Genetics Studies

A Federal law, called the Genetic Information Nondiscrimination Act (GINA), effective May 21, 2010, generally makes it illegal for health insurance companies, group health plans, and most employers to discriminate against you based on your genetic information. This law generally will protect you in the following ways:

- Health insurance companies and group health plans may not request your genetic information that we get from this research.
- Health insurance companies and group health plans may not use your genetic information when making decisions regarding your eligibility or premiums.
- Employers with 15 or more employees may not use your genetic information that we get from this research when making a decision to hire, promote, or fire you or when setting the terms of your employment.

Be aware that this Federal law does not protect you against genetic discrimination by companies that sell life insurance, disability insurance, or long-term care insurance.

The results of the analysis of your child's DNA done as part of this study will not be given to you.

Your child's DNA sample will be identified by a code number, and all other identifying information will be removed. Dr. Lewis will keep a separate code sheet which links the DNA sample code number with your identity.

Your child's sample may be stored indefinitely. If in the future, you change your mind and would prefer not to have your child's DNA used for research you can contact Dr. Lewis at (216) 368-4674 and request that any existing samples linked to you be destroyed.

The information obtained as a result of your participation in this research wi
record. Information from which your child may be personally identified will be maintained in a confidential, locked file at CWRU on the 4th floor of the Cleveland Hearing and Speech Center and will not be disclosed to third parties except with your permission or as may be required by law.

When your child turns 18 years of age, he/she would need to consent as an adult for his/her DNA material to continue to be used.

Your child can participate in this research study even if you do not want to have his/her saliva sample taken for DNA (gene) studies. Please indicate below your choice.

❑ Yes, I want to have my child donate a sample for the DNA (gene) studies.

❑ No, I do not want my child to participate in the DNA (gene) studies.

Your DNA (genes) or your cells that can be used to make your DNA will be stored for research purposes. Please check one of the following options telling us how your DNA samples may be used.

- ❑ My samples may be used for this project only. Do not use them for any other project and do not contact me again for permission.

- ❑ My samples may be used for this project only and for other projects with my permission. If my samples could be used for another project, contact me to ask my permission.

- ❑ My samples may be used for any scientific purposes involving this or any other project. Do not contact me again for permission.

You have the right to withdraw from this study. If you choose to withdraw no further studies will be completed on the samples that your child has contributed. You may withdraw your child's DNA sample from the study by contacting Dr. Barbara Lewis at (216) 368-4674 or Gabrielle Miller at (781)-640-1044.

Termination of Participation

Your participation in this study may be discontinued by the investigator without your consent if your child does not meet inclusion criteria for the study.

Confidentiality

The records of this research will be kept confidential. Any time information is collected, there is a potential risk for loss of confidentiality. Every effort will be made to keep your information confidential; however, this cannot be guaranteed.

In any sort of report we might publish, we will not include any information that will make it possible to identify a participant. Research records will be kept in a locked file and access will be limited to the researchers, and the University review board responsible for protecting human participants

Subject Identifiable Data

Some information that identifies you will be removed and replaced with a code. A list linking the code and to your identifiable information will be kept separate from the research data in a secure location. Access to files will be restricted to key study personnel and supervised by the principal investigator(s) of the study.

Other information that can identify you such as your signed consent form and the Developmental Questionnaire will be kept in a separate locked file cabinet without a link to the research data.

Data Storage

Research data will be maintained in a secure location at CWRU. Only authorized individuals will have access to it. Research data will be stored electronically in an encrypted file and is password protected.

The audio recordings that can identify you will be:

- Stored in a secure location and,
- Transcribed and erased as soon as possible.

Data Retention

The researchers intend to keep the research data:

- Until the research is published and/or presented;
- For approximately 10 years

Contacts and Questions

The researchers conducting this study are Barbara Lewis and Gabrielle Miller. You may ask any questions you have now. If you have any additional questions, concerns or complaints about the study, you may contact them at (216) 368-4674.

If the researchers cannot be reached, or if you would like to talk to someone other than the researcher(s) about; (1) questions, concerns or complaints regarding this study, (2) research participant rights, (3) research-related injuries, or (4) other human subjects issues, please contact Case Western Reserve University's Institutional Review Board at (216) 368-4514 or write: Case Western Reserve University; Institutional Review Board; 10900 Euclid Ave.; Cleveland, OH 44106-7230.
You will be given a copy of this form for your records.

Permission to Record

Audio or video recording is an integral part of the study and if you do do not wish to have your child audio recorded, they should not participate in the study.

Statement of Consent

Your signature below certifies the following:

- You have read (or been read) the information provided above.
- You have received answers to all of your questions and have been told who to call if you have any more questions.
- You have freely decided to participate in this research.
- You understand that you are not giving up any of your legal rights.

_____
Printed Name of Participant's Parent:

_____ Date:_____
Signature of Participant's Parent:

_____
Printed Name of Legally Authorized Representative

_____ Date:_____
Signature of Legally Authorized Representative

_____ Date:_____
Signature of Person Obtaining Consent

Appendix B - Assent

Version: 08/2013

CASE WESTERN RESERVE UNIVERSITY
INFORMED ASSENT DOCUMENT
Speech and Reading Study

Participating in this study is totally voluntary. I am going to read this information to you. Feel free to ask questions about anything that you do not understand before deciding if you want to be in the study. A researcher listed below will be around to answer your questions.

WHY ARE YOU HERE?
The researchers want to tell you about a research study looking at speech, language and reading. They want to see if you would like to be in this study.

WHY ARE THEY DOING THIS STUDY?
Dr. Barbara Lewis and Gabrielle Miller, and some other researchers are doing this study to learn more about what helps kids to be good readers. There will be about 40 kids in this study.

WHAT WILL HAPPEN TO YOU?
These things will happen if you want to be in the study:

1. You will be asked to see a language and reading teacher and you will be asked to stay for about four hours.
2. We will ask you to take some speech, language, reading and other tests like those that you do in school.
3. You will be asked to wear headphones and have your hearing screened
4. You will be audio recorded.
5. If you and your parents agree, we will ask you to spit in a cup.

WILL THE STUDY HURT?
You may get tired from taking the speech, language tests and reading. You may take a break when you need to.

WILL YOU GET BETTER IF YOU ARE IN THE STUDY?
You will not get better but this study might find out things that will help other children with speech, language or reading problems some day.

HOW WILL YOUR INFORMATION BE PROTECTED?
The information collected about you during this study will be kept safely locked up. Nobody will know it except the people doing the research.

The study information about your speech, language and reading will be given to your parents.

WHAT IF YOU HAVE ANY QUESTIONS?
You can ask questions any time. You can ask now or you can ask later. You can talk to the researchers, your mom and dad, or you can talk to someone else. You may ask any questions you have now. If you have any additional questions, concerns or complaints about the study, you may contact the researchers at 781-640-1044.

If the researchers cannot be reached, or if you would like to talk to someone other than the researcher(s) about; (1) questions, concerns or complaints, (2) your rights, (3) research-related injuries, or (4) other issues, please contact Case Western Reserve University's Institutional Review Board at (216) 368-6925 or write: Case Western Reserve University; Institutional Review Board; 10900 Euclid Ave.; Cleveland, OH 44106-7230.

DO YOU HAVE TO BE IN THE STUDY?
You do not have to be in the study. No one will be mad at you if you don't want to do this. If you don't want to be in this study, you just have to tell the researchers. You can say yes now and change your mind later. It is up to you to decide.

If you decide to be in the study, please write your name below.

You can change your mind and stop being part of it at any time. All you have to do is tell the person in charge.

You will be given a copy of this paper to keep.

_____ Age: _____
Printed Name of Participant

_____ Date: _____
Signature of Participant

_____
Printed Name of Researcher/Person Obtaining Assent

_____ Date: _____
Signature of Researcher/Person Obtaining Assent

Appendix C - Developmental Questionnaire

SPEECH-LANGUAGE AND READING QUESTIONNAIRE

I. BASIC INFORMATION

Name: _____ DOB: _____

Address: _____ Age: _____

_____ Grade: _____

Phone: _____ Email: _____

Gender: _____ Handedness: _____

Parents' names: _____

OCCUPATION OF PARENTS

Mother: _____ Father: _____

Person completing questionnaire: _____

Relationship to the child: _____

Mother's ethnicity: ☐ Hispanic or Latino ☐ Non-Hispanic or Latino

Mother's race: ☐ CAUCASIAN ☐ AFRICAN-AMERICAN ☐ INDIAN ☐ MIDDLE EASTERN
☐ EAST ASIAN ☐ PACIFIC ISLANDER ☐ NATIVE AMERICAN

Father's ethnicity: ☐ Hispanic or Latino ☐ Non-Hispanic or Latino

Father's race: ☐ CAUCASIAN ☐ AFRICAN-AMERICAN ☐ INDIAN ☐ MIDDLE EASTERN
☐ EAST ASIAN ☐ PACIFIC ISLANDER ☐ NATIVE AMERICAN

Languages spoken at home: _____

II. PRENATAL HISTORY

Please describe any medical attention that was required during pregnancy:

_____

_____

_____

III. DEVELOPMENTAL HISTORY

Child's birth weight: _____ Length: _____

Did your child have any feeding issues as an infant or toddler? Describe:

_____

_____

_____

Ages at which the following developmental milestones were achieved:

Held head up: _____ Stood: _____

Sat: _____ Walked: _____

Crawled: _____ First words: _____

IV. MEDICAL HISTORY

- ☐ Adenoidectomy Age: _____
- ☐ Allergies Age: _____
- ☐ Asthma/Bronchitis Age: _____
- ☐ Body dyspraxia (clumsiness) Age: _____
- ☐ Chronic Colds Age: _____

- ☐ Dental Problems Age: _____
- ☐ Drooling Age: _____
- ☐ Ear Infections Age: _____
- ☐ Encephalitis Age: _____
- ☐ Epilepsy Age: _____
- ☐ Fine Motor Issues Age: _____
- ☐ Gross Motor Issues Age: _____
- ☐ Head Injuries Age: _____
- ☐ Hearing Aids Age: _____
- ☐ Heart Condition Age: _____
- ☐ Lead Exposure Age: _____
- ☐ Meningitis Age: _____
- ☐ Neurological problems Age: _____
- ☐ Orthodontia Age: _____
- ☐ Seizures Age: _____
- ☐ Tonsillectomy Age: _____
- ☐ Tonsillitis Age: _____
- ☐ Tubes in ears Age: _____
- ☐ Visual deficits Age: _____

Comments:_____

_____

_____

_____

Describe any illnesses, accidents, surgeries or hospitalizations that your child has had.

_____

_____

_____

V. SPEECH-LANGUAGE DEVELOPMENT

Reduced vocal play or babbling	YES__ NO__
Reduced imitation in infancy	YES__ NO__
Family history of speech disorders	YES__ NO__
Family history of language disorders	YES__ NO__
Family history of reading difficulties	YES__ NO__
Delayed language onset	YES__ NO__
Slow progress Speech therapy	YES__ NO__
Diagnosis of Dysarthria	YES__ NO__
Reduced repertoire of sounds	YES__ NO__

Comments:

_____

_____

_____

_____

1. Does your child currently have difficulty producing any sounds? Please describe:

_____

_____

2. Does your child have difficulty comprehending age appropriate vocabulary?

_____

_____

_____

3. Does your child have difficulty following spoken directions?

_____

_____

_____

4. Does your child have difficulty processing verbal information (e.g., stories, conversation)? _____

_____

_____

5. Does your child have difficulty explaining ideas? (e.g., trouble finding the right word, difficulty telling a story in order, using excessive filler words such as "um", "uh" etc.)

_____

_____

_____

6. How would you rate your child's speaking voice? (e.g., normal quality, too soft, too loud, hypernasal, stuffed up sounding, hoarse)?

_____

_____

V. EDUCATIONAL HISTORY

1. Did your child receive early intervention services as an infant or toddler (i.e., OT, PT, speech therapy)?

_____

_____

_____

2. Did your child receive any special education services in preschool (i.e., OT, PT, speech therapy)? _____

_____

_____

3. Did your child receive any special education services in elementary school (OT, PT, speech therapy, reading, math etc.)?

_____

_____

_____

4. Is your child currently receiving IEP services (please describe)?

_____

_____

_____

_____

5. Does your child currently receive tutoring outside of school for reading or math (please describe)?

6. If applicable, describe your child's difficulty with reading and/or spelling?

7. Does anyone in your immediate family (i.e., you, your spouse, siblings of participant) have a history of speech, language or academic difficulties? (please describe)

Chapter 6. References

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