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Motor Performance in Children with Childhood Apraxia of Speech and Speech Sound Disorders

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Abstract
Purpose: This study sought to determine if (a) children with childhood apraxia of speech (CAS), other speech sound disorders (SSDs), and typical development (TD) would perform differently on a standardized motor assessment and (b) whether comorbid language impairment would impact group differences.
Method: Speech, language, and motor abilities were assessed in children with CAS (n = 10), SSD (n = 16), and TD (n = 14) between the ages of 43 and 105 months. Motor skills were evaluated using the Movement Assessment Battery for Children-Second Edition (Henderson, Sugden, & Barnett, 2007), a behavioral assessment that is sensitive in identifying fine/gross motor impairments in children with a range of motor and learning abilities. Data were reanalyzed after reclassifying children by language ability.

Results: The CAS group performed below the normal limit on all components of the motor assessment and more poorly than the TD and SSD groups on Aiming and Catching and Balance. When children were reclassified by language ability, the comorbid CAS + language impairment group performed worse than the SSD-only and TD groups on Manual Dexterity and Balance and worse than the TD group on Aiming and Catching; all 7 children with CAS + language impairment evidenced performance in the disordered range compared to 1 of 3 children in the CAS-only group and 2 of 6 children in the SSD + language impairment group.

Conclusions: Children with CAS + language impairment appear to be at an increased risk for motor impairments, which may negatively impact social, academic, and vocational outcomes; referrals for motor screenings/assessments should be considered. Findings may suggest a higher order deficit that mediates cognitive-linguistic and motor impairments in this population.

Introduction

Childhood apraxia of speech (CAS) is a neurological pediatric speech disorder characterized by poor planning and/or programming of speech sound sequences (American Speech-Language-Hearing Association [ASHA], 2007). Children with CAS evidence a constellation of symptoms including, but not limited to, inconsistent speech sound production, vowel errors, difficulty with co-articulatory transitions, and prosodic disturbances (ASHA, 2007; Iuzzini-Seigel, Hogan, & Green, 2017; Iuzzini-Seigel & Murray, 2017); in addition, poor response to intervention is common, and many will participate in speech treatment throughout childhood and into adolescence (ASHA, 2007; Iuzzini & Forrest, 2010; Maas & Farinella, 2012; Murray & Iuzzini-Seigel, 2017). Children in this population are not only negatively affected by atypical speech production but are also at an increased risk for language impairments and fine and gross motor deficits (Teverovsky, Bickel, & Feldman, 2009; Tükel, Björelius, Henningsson, McAllister, & Eliasson, 2015; Zuk, Iuzzini-Seigel, Cabbage, Green, & Hogan, 2018), thereby increasing their risk for academic, social, and vocational challenges (Lewis et al., 2004).

Depending on the severity of a child's motor impairment, fine and gross motor deficits may continue into adolescence and adulthood and impact mobility, self-feeding, self-care, writing, and participation in physical and athletic activities (Cantell, Smyth, & Ahonen, 1994; Hellgren, Gillberg, Bågenholm, & Gillberg, 1994; L. T. Miller, Missiuna, Macnab, Malloy-Miller, & Polatajko, 2001). Physical limitations may also further compound the social consequences of communication impairments and be associated with poorer self-esteem, bullying, and other psychosocial and psychiatric issues (Bouffard, Watkinson, Thompson, Dunn, & Romanow, 1996; Cantell et al., 1994; Cousins & Smyth, 2003; Geuze & Börger, 1993; Hellgren et al., 1994).
Equivocal Findings on Motor Function in Children With CAS

The research on generalized motor function in children with symptoms and/or diagnosis of CAS is limited, and results are equivocal (Bradford & Dodd, 1996; Dewey, Roy, Square-Storer, & Hayden, 1988; Gretz, 2013; Newmeyer et al., 2007; Potter, Nievergelt, & Shriberg, 2013; Tükel et al., 2015). Apraxia Kids (formerly the Childhood Apraxia of Speech Association of North America) reports that responses from a parent survey indicate that approximately 50% of children with CAS have a history of physical and/or occupational therapy (Gretz, 2013). Similarly, Dewey et al. investigated sequencing of oral and limb gestures in children with poor sequencing of consonants and vowels—symptoms of CAS (Dewey et al., 1988)—and found that those with speech sequencing difficulties also demonstrated difficulty with oral and limb sequencing. Potter et al. (2013) examined movement in children with galactosemia, a disorder that prevents metabolism of the milk sugar "galactose" and in which high rates of cognitive-linguistic, speech, and motor impairments are reported to occur (Shriberg, Potter, & Strand, 2011)—nearly 25% of children with galactosemia are reported to have CAS (Shriberg et al., 2011). Potter et al. found that 21 of 32 participants with galactosemia and comorbid speech disorders evidenced motor performance below the 10th percentile on the Movement Assessment Battery for Children (Movement ABC; Henderson & Sugden, 1992), indicating motor impairments among the majority of participants with this diagnosis. The Movement ABC, described in greater detail below, is a test of fine and gross motor abilities that provides valid assessment of children with a range of cognitive abilities and that can be used to identify even mild motor impairments. Bradford and Dodd (1996) used the Bruininks-Oseretsky Test of Motor Proficiency (BOT; Bruininks, 1978) to investigate fine motor performance in children with typical development (TD), developmental verbal dyspraxia (aka CAS), and other speech sound disorders (SSDs), including consistent deviant speech disorder, speech delay, and inconsistent speech disorder. Results revealed fine motor deficits in timed tasks among children with CAS and inconsistent speech disorder. Bradford and Dodd suggested that, for children with inconsistent speech disorder, these fine motor deficits could relate to a difficulty in incorporating timing into complex motor plans, whereas those with CAS may evidence a generalized deficit in motor planning.

In contrast, Newmeyer et al. (2007) used the Peabody Developmental Motor Scales-Second Edition (PDS-2; Folio & Fewell, 2000) to investigate movement in 32 preschoolage children with severe SSD. Results showed that difficulty with oral motor imitation (i.e., a sign of oral motor apraxia but not necessarily CAS) was associated with poor fine motor performance and fine motor scores in the disordered range whereas speech deficits were not. Newmeyer et al. attributed this relation to a possible issue with the mirror neuron system, which could negatively impact imitated motor movements, including oral function and fine motor tasks. Tükel et al. (2015) used the BOT to assess motor function in 18 children with CAS and found a different result. On average, participants scored within 1 SD of the mean on the BOT, indicating normal motor performance. It should be noted that Tükel et al.’s sample was restricted to children with limited language deficits and no history of physical or occupational therapy, likely limiting the severity and complexity of the participants and thereby reducing generalization of their findings. In fact, recent work that controlled for language ability in children with CAS showed that some deficits (i.e., speech perception deficits) may only be evidenced by children with CAS and comorbid language impairment and not by those with CAS and typical
language ability (e.g., Zuk et al., 2018). Consequently, language ability is an important variable to consider and control for when working with children in this population.

Equivocal findings in these studies may be due in part to differences in the sensitivity of the motor instruments that were utilized. For instance, investigation of convergence validity between the PDS-2 and Movement ABC revealed that the Movement ABC was more sensitive in detecting mild-moderate deficits compared to the PDS-2 (Van Waelvelde, Peersman, Lenoir, & Engelsman, 2007). Differing results across studies may also be explained by differences in CAS diagnostic criteria, such that participant samples may have varied widely across studies. There is no commonly agreed-upon validated list of criteria that is used to differentiate CAS from SSD. Consequently, researchers and clinicians vary in the features they use to make this diagnosis and on the procedures they use to elicit the features. Whereas some may include nonspeech oral sequencing measures during the differential diagnostic process (Lewis et al., 2004; Parsons, Cox, & Reed, 1988; Smith, Marquardt, Cannito, & Davis, 1994), others rely entirely on speech features (Iuzzini-Seigel & Murray, 2017; Iuzzini-Seigel et al., 2017; Maas & Mailend, 2017; Moss & Grigos, 2012; Shriberg et al., 2011). Finally, equivocal motor results may be explained by varying language abilities among participants within and across these studies, similar to what has been found with speech perception (Froud & Khamis-Dakwar, 2012; Groenen, Maassen, Crul, & Thoonen, 1996; Maassen, Groenen, & Crul, 2003; Nijland, 2009; Zuk et al., 2018).

The extant literature shows that children with language impairment tend to perform more poorly than typically developing children on motor tasks (Bishop, 2002, 2005; Hill, 2001; Kent, 1984), although sometimes still within the normal range (Zelaznik & Goffman, 2010). This cooccurrence is not surprising given the interaction between the motor and cognitive-linguistic systems (Floel, Ellger, Breitenstein, & Knecht, 2003; Goffman, 2004, 2010; Iuzzini-Seigel, Hogan, Rong, & Green, 2015; Walsh, Smith, & Weber-Fox, 2006; Zelaznik & Goffman, 2010) and the overlap of neural substrates that serve these systems (Jäncke, Siegenthaler, Preis, & Steinmetz, 2007; Kent, 2004; Leiner, Leiner, & Dow, 1991, 1994). For instance, cerebellar dysfunction could yield co-occurring speech, motor, and cognitive-linguistic deficits (Bracke-Tolkmitt et al., 1989; Leiner et al., 1991, 1994). Poor motor performance among children with language impairments (e.g., Powell & Bishop, 1992; Zelaznik & Goffman, 2010) does not suggest that language impairment underlies motor impairments, but rather, it signals the possibility of a higher order mechanism that mediates cognitive-linguistic and motor performance. For instance, co-occurring language and fine/gross motor deficits could reflect poor procedural learning ability (e.g., Nicolson & Fawcett, 2007), difficulty in integrating sensory information (e.g., Tallal, Miller, & Fitch, 1993), or reduced information-processing capacity (e.g., C. A. Miller, Kail, Leonard, & Tomblin, 2001). The procedural learning deficit hypothesis has been used to explain speech, language, and motor comorbidities in children with specific language impairment (Hedenius et al., 2011; Lum, Ullman, & Conti-Ramsden, 2013; Nicolson & Fawcett, 2007). The procedural learning system is the mechanism by which we learn patterns (e.g., phonological patterns, grammatical rules) without being explicitly taught. Practice and multiple repetitions help patterns to become stored in the procedural memory system and lead to automaticity, such that these patterns are produced faster and more effortlessly over time. Consequently, a procedural learning deficit can theoretically lead to impairments in both cognitive-linguistic and motor domains (Nicolson & Fawcett, 2007), including comorbidity of attention, speech, language, and motor impairments as commonly observed in at least a subset of children with CAS. There is a gap in our knowledge as to what extent children with CAS, with and without language
impairments, evidence fine and gross motor impairments—information that is essential to inform the underlying nature and anatomical underpinnings of CAS, as well as guide clinical referrals, and inform treatment planning and complete care of individuals in this population.

Purpose and Research Questions
The current study investigated fine and gross motor ability in children with idiopathic CAS and control groups of children with TD and non-CAS SSD. The Movement Assessment Battery for Children—Second Edition (Movement ABC-2; Henderson, Sugden, & Barnett, 2007), which has good sensitivity and validity for detecting even mild motor deficits in children with a range of learning and cognitive abilities (Henderson & Sugden, 1992; Lam & Henderson, 1987; Spandet al., 1999; Sugden & Wann, 1987), was used to assess fine and gross motor abilities. This test uses a variety of tasks to assess Manual Dexterity, Aiming and Catching, and Balance, as described in detail below. Our well-established diagnostic protocol for differentially diagnosing CAS and SSD (Iuzzini-Seigel et al., 2017) was used to ensure internal validity and encourage replication. We posited that children with CAS would evidence deficits in motor tasks and, in particular, fine motor tasks (i.e., Manual Dexterity), as these are most similar to the fine-grained and precise movements required for speech, whereas those with SSD or TD would evidence good motor performance across tasks. We also hypothesized that children with CAS would evidence poor Balance and Aiming and Catching skills, as these rely on proprioception and graded movements, much in the way that vowel production—an area of particular challenge for children with CAS (Davis, Jacks, & Marquardt, 2005; Nijland et al., 2002; Pollock & Hall, 1991)—does. Furthermore, we anticipated that fine and gross motor performance would be correlated with speech and language measures, such that children with more severe speech and language impairments would perform more poorly on motor assessments than those with better speech and language abilities.

Finally, data were reanalyzed after reassigning children to groups based on language performance. Based on the extant literature investigating motor performance in children with language impairments, we posited that children with comorbid speech and language impairments (CAS + LI and SSD + LI) would perform more poorly than peers with typical language abilities (i.e., CAS-only, SSD-only, TD). Additionally, we hypothesized and that children with CAS + LI would have poorer motor abilities than those with SSD + LI.

Method
Ten children with CAS and 30 age-matched controls (14 with TD, 16 with SSD) participated in this study. Children ranged in age between 43 and 105 months (M =73 months, SD =15, Mdn = 75 months). Exclusionary criteria included oromotor weakness or orofacial dysmorphology, cognitive impairments that prevented participation in experimental procedures and tasks, and hearing impairment. All procedures were approved by the Marquette University Institutional Review Board. All participants completed a series of communication, cognitive, and motor assessments, including the Sounds-in-Words subtest of the Goldman-Fristoe Test of Articulation—Third Edition (GFTA-3; Goldman & Fristoe, 2015), Receptive and Expressive Language components of the Clinical Evaluation of Language Fundamentals—Fifth Edition (CELF-5; Wiig, Secord, & Semel, 2013) or Clinical Evaluation of Language Fundamentals Preschool—Second Edition (CELF Preschool-2; Wiig, Secord, & Semel, 2004), the nonverbal components of the Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003), and the Movement ABC-2 (Henderson et al., 2007). Three participants completed the Test of Integrated
Language and Literacy Skills (TILLS; Nelson, Plante, Helm-Estabrooks, & Hotz, 2016) instead of the CELF. The TILLS does not yield separate receptive and expressive language scores, and consequently, we report a composite core abilities score for these children in Table 1, which reports speech and language data for individual participants. All participants passed a pure-tone hearing screening for the frequencies of 1000, 2000, and 4000 Hz at 20 dB and 500 Hz at 25 dB. Participants also completed the oral mechanism structure and oral function components of the Robbins and Klee (1987) assessment. Oral function tasks were elicited using verbal prompts and models where needed; tasks assessed functions such as lip and tongue protrusion, lip seal, tongue elevation, and anterior-posterior tongue movement. Speech was not assessed as part of this oral function assessment. All testing was completed over a series of three or four sessions; sessions were 90–120 min each, with breaks given as needed. Sessions were led by undergraduate and graduate students of speech pathology who were trained as research assistants.

**Group Assignment**

Children were assigned to groups based on standardized and custom assessments using a well-established protocol in our lab (Iuzzini-Seigel et al., 2017; Iuzzini-Seigel & Murray, 2017; Zuk et al., 2018). A licensed speech-language pathologist with extensive experience and training on rating CAS features listened to all speech samples and assigned children to groups. This rater was blinded to any previous differential diagnosis participants may have had. Next, a speech pathology student who had completed the rigorous feature rating training in our lab rerated CAS features for 15% of the participants divided across groups. Interrater reliability for feature ratings was 92%. See Table 1 for speech and language scores by participant.

CAS features used to determine group assignment included vowel errors, consonant distortions, stress errors, syllable segregation, groping, intrusive schwa, voicing errors, slow rate, increased difficulty with multisyllabic words, resonance or nasality disturbance, difficulty in achieving initial articulatory configurations or transitional movement gestures, and inconsistency. See Iuzzini-Seigel et al. (2017) or Iuzzini-Seigel and Murray (2017) for explicit operational definitions for each feature and Iuzzini and Forrest (2010) for further explanation of the inconsistency measure. CAS features were rated across the following speech tasks: two administrations of the Sounds-in-Words subtest on the GFTA-3, a customized list that elicits words (Iuzzini-Seigel et al., 2017; Shriberg, Jakielski, & Strand, 2010) and nonwords (Nelson et al., 2016) of varying lengths and complexity levels produced in isolation, and a speech sample elicited by the Park Play picture (Patel & Connaghan, 2014). The build upon word list contains stimuli of increasing length, such that triads contain the same root word (e.g., lay, lady, ladybug) and help to determine if a child is having increased difficulty with multisyllabic words compared with monosyllabic targets. The multisyllabic word list contains challenging words that range from three to four syllables (e.g., sympathize). The nonwords (Nelson et al., 2016) contain mono- and multisyllabic exemplars ranging from one to five syllables. These lists were prerecorded by Midwestern talkers, and stimuli were presented via sound field at a comfortable listening level. The build upon word and nonword lists were each presented once, and the multisyllabic word list was presented twice with a different task in between the two administrations.

Inconsistency was measured at the phonemic level and was determined using the Inconsistency Severity Percentage (Iuzzini & Forrest, 2010; Iuzzini-Seigel et al., 2017):
\[
\Sigma((\text{number of different error types} - 1 \text{ for each phoneme})/
\text{2(total number of target opportunities)}) \times 100. \tag{1}
\]

Inconsistency severity percentages of 18% or higher were considered positive for inconsistency (Iuzzini, 2012; Iuzzini-Seigel et al., 2017). Participants were assigned to the CAS group (\(n = 10\)) if they evidenced a GFTA-3 standard score of < 85, inconsistency, and an average of five or more CAS features (Iuzzini-Seigel et al., 2017; Iuzzini-Seigel & Murray, 2017). Inclusion in the SSD group (\(n = 16\)) was based on a GFTA-3 standard score of < 85, an inconsistency severity percentage of < 18%, < 5/11 CAS features, and no previous diagnosis of CAS or history of treatment for CAS; the last criterion was specified to prevent inclusion of children in the SSD group who had resolved CAS symptoms. Participants were assigned to the TD group (\(n = 14\)) based on a GFTA-3 standard score of > 85, < 5/11 CAS features, an inconsistency severity percentage of < 18%, typical language, and no history of speech or language treatment; the last criterion was to exclude children who had remediated speech or language deficits. For the majority of participants in the typically developing group, typical language was based on a Receptive Language score of > 85 on the CELF Preschool-2 or CELF-5. Although it was not a criterion, all children in the TD group who completed a CELF assessment also had Expressive Language scores in the normal range (> 85). The cut score to diagnose language/literacy disorders on the TILLS is 24 for children aged 72-95 months and 34 for children aged 96-143 months; consequently, typical language abilities were indicated for all three children (two with SSD, one with TD) who completed this testing instrument. Because the TILLS does not offer receptive and expressive language composite scores that are comparable to the CELF measures, the language scores for these three children were used for group assignment but omitted from statistical analyses.

**Differential Diagnosis of Challenging Cases**

One child (Participant 009) evidenced 5.6 CAS features but an inconsistency severity percentage of 5.9% on productions from the GFTA-3. He evidenced moderate-high intelligibility but a severe prosodic disturbance. Consequently, he did not neatly meet criteria for assignment to either the CAS or SSD group. It should be noted that this child, who was 73 months of age, had previously participated in speech treatment, and previous research shows that children with CAS may decrease phonemic inconsistency following treatment, such that they pattern more like children with phonological disorder rather than children with CAS (Iuzzini & Forrest, 2010). To further determine his differential diagnosis, we aimed to tax his system and assessed lexical inconsistency across two productions of the multisyllabic word list, which contains challenging words. Participant 009 was inconsistent on 50% of targets. Given the number of CAS features, characteristic severe prosodic disturbance, and presence of lexical inconsistency, this child was assigned to the CAS group.

A second child (Participant 025) evidenced a GFTA-3 standard score of 95, 3.9 CAS features, and lower accuracy and intelligibility in connected speech and other challenging contexts. He also reported a history of speech treatment. Given this profile, this child was assigned to the SSD group.

**Movement Assessment**

The Movement ABC-2 was used to assess motor competency at the behavioral level. According to the manual, this test "may be used by professionals with a variety of backgrounds and training from both
health and education” (Henderson et al., 2007, p. 6). The Movement ABC-2 takes 20-30 min to complete, and data yield three component scores: Aiming and Catching, Balance, and Manual Dexterity. The test has been shown to be effective in assessing fine and gross motor abilities in children with a range of language and cognitive abilities (Henderson & Sugden, 1992; Spano et al., 1999; Sugden & Wann, 1987). The Aiming and Catching component, which assesses gross motor ability, consists of catching a beanbag or tennis ball and throwing a beanbag onto a mat. The Balance component, which assesses this foundational skill required for gross motor competence, consists of balancing on one leg, walking with heels raised or walking along a line using a heel-to-toe strategy, and jumping or hopping. The Manual Dexterity component, which assesses fine motor ability, is composed of three tasks including drawing trails, threading beads or threading a lace in and out of a board with holes on it, and either placing coins into a coin slot (e.g., as in a piggy bank) or placing mushroom-shaped pegs onto a plastic pegboard; this assessment tests both the child’s dominant and nondominant hand. For the drawing trails subtest, children are presented with a line drawing of a bicycle trail and are asked to draw a single continuous line from one end of the trail to the other, making sure to stay within the boundaries of the trail; for older children, this trail has sharper angles, making it more challenging.

Tests were administered and scored by trained research assistants in accordance with procedures documented in the manual. Each task included a demonstration and a practice phase. Each component (i.e., Manual Dexterity, Balance, Aiming and Catching) has a mean standard score of 10 and an SD of 3, such that scores below 7 indicate performance below the normal limit and performance with scores of 5 or below indicates significant movement difficulty that will likely require intervention by a physical and/or occupational therapist (Henderson et al., 2007).

Data Analysis
Statistical assumptions were assessed for each variable, and nonparametric tests were used where necessary. Analyses of variance (ANOVAs) or Kruskal-Wallis tests were used to detect group differences for age, nonverbal IQ, articulation, oral mechanism structure and oral function scores, Expressive Language, Receptive Language, Manual Dexterity, Aiming and Catching, and Balance. Post hoc t tests, Games-Howell tests (if assumption of homoscedasticity was not met), or Mann-Whitney U tests were used to detect pairwise differences. The Bonferroni adjustment to control for familywise error rate was used where indicated. Effect sizes were calculated for group differences detected for Movement ABC-2 components. Planned Spearman’s rho correlations were used to determine relations between speech, language, oral function, and motor variables.

After data were analyzed by group, participants in the CAS and SSD groups were reclassified on the basis of CELF Preschool-2 or CELF-5 Receptive Language Index standard scores. The Receptive Language Index for the CELF Preschool-2 (i.e., used for children 3-6 years of age) reflects performance on the Sentence Structure, Concepts and Following Directions, and Basic Concepts or Word Classes subtests. For the CELF-5 (i.e., used for children older than 6 years of age), the Receptive Language Index reflects performance on the Sentence Comprehension, Word Classes, and Following Directions subtests. Those with Receptive Language Index scores of 85 or below were assigned to the CAS + LI or SSD + LI groups. We have often determined the presence of language impairment using a cutoff of 85 on the Core Language composite from the CELF Preschool-2 or CELF-5 (Centanni, Green, IuzziniSeigel, Bartlett, & Hogan, 2015; Centanni, Sanmann, et al., 2015; Iuzzini-Seigel, Hogan, Guarino, & Green, 2015; Iuzzini-Seigel et al., 2017; Zuk et al., 2018). This test cutoff yields adequate sensitivity and specificity in
accurately identifying language disorder in children with true language impairments and not misidentifying language disorder in children with typical language development (Spaulding, Plante, & Farinella, 2006); however, it is heavily weighted with expressive language items that can be challenging to score for children with severe speech deficits and compromise the internal validity of this measure. In the current study, we used a cutoff of 85 on the Receptive Language Index from the CELF Preschool-2 and CELF-5 to differentiate groups. Of the 10 participants who met criteria for the CAS group, three exhibited CAS with normal receptive language (CAS only) and seven exhibited CAS with comorbid language impairment (CAS + LI). Of the 16 participants with SSD, 10 had typical language (SSD only) and six had comorbid language impairment (SSD + LI). Because typically developing language was required for initial assignment to the TD group, all children in the TD group maintained their initial group membership in the TD group. Due to unequal and small group sizes, Kruskal-Wallis and Mann-Whitney U tests were used to detect differences in motor performance between groups recategorized by language performance.

Results

Demographics

ANOVAs or Kruskal-Wallis tests were used to detect group differences in demographic, speech, language, oral mechanism, and cognitive variables. See Table 2 for a summary of participant data by group, including notation of statistically significant differences between groups. No statistically significant differences were detected between groups for age, nonverbal IQ, or oral mechanism structure scores. As expected, due to the basis for group assignments, ANOVAs revealed a main effect of group for CAS features, $F(2, 37) = 85.642$, $p < .001$, wherein children with CAS evidenced more features ($p < .001$) than other groups and the SSD group evidenced more features ($p < .001$) than the TD group. On average, children with CAS evidenced six features, while those with SSD and TD evidenced three and two features, respectively.

A group effect was also revealed for expressive language, $F(2, 34) = 18.448$, $p < .001$, wherein the CAS group performed more poorly compared to the other groups ($p < .001$). Kruskal-Wallis tests revealed group differences ($p < .003$) in articulation based on GFTA-3 standard scores, inconsistency severity percentages, receptive language, and oral function. Mann-Whitney U tests revealed that the CAS group evidenced lower scores on these measures than the TD and SSD groups; likewise, the SSD group scored lower than the TD group on articulation, inconsistency, and receptive language. The three children who completed the TILLS did not have separate receptive and expressive language scores, and therefore, their language scores were considered missing data in descriptive measures and statistical comparisons.

Fine and Gross Motor Performance

Parent responses on case history forms reported that two of 10 children in the CAS group had a history of attending physical and occupational therapy and one additional child in this group had undergone evaluation for physical and occupational therapy but did not participate in therapy. One child in the SSD group reported a history of attending occupational therapy. An additional six parents from the CAS group, one from the TD group, and three from the SSD group reported concerns about their child’s fine
and/or gross motor abilities, although these children had not been seen by physical or occupational therapists.

ANOVA detected group differences in Aiming and Catching, $F(2, 37) = 9.099$, $p = .001$, $np^2 = .327$, power $= .927$, and Balance, $F(2, 37) = 10.575$, $p < .001$, $np^2 = .354$, power $= .954$. No group differences were revealed for Manual Dexterity. Bonferroni-adjusted (.05/3) pairwise comparisons resulted in setting an adjusted significance level of .016. Post hoc t tests revealed that, for Aiming and Catching, the CAS group performed more poorly than children with SSD ($p = .004$) and TD ($p < .001$). Likewise, for the Balance component, the CAS group performed more poorly than the SSD group ($p = .001$) and the TD group ($p < .001$). See Figure 1 for motor performance by group on the Movement ABC-2.

Total performance scores on the Movement ABC-2 can be used to indicate whether a child demonstrates significant movement difficulty or is at risk for movement difficulty and should be monitored. Eight of 10 children with CAS, three of 14 children with TD, and three of 16 children with SSD evidenced a total performance standard score of 5 or below, indicating significant movement difficulty. In addition, two of 16 children with SSD had a total performance standard score of 6, indicating they were at risk for movement difficulty and should be monitored. Of the 14 children whose performance indicated significant movement difficulty, eight parents had reported concern about their child's motor skills on the case history form, yet only four of these children had previously undergone a physical or occupational therapy evaluation and only three of four had participated in any type of movement therapy.

Relation Between Oral Motor Function and Motor Abilities
Spearman’s rho correlations were conducted between oral motor function scores and each of the Movement ABC-2 components. Findings revealed a significant moderate positive correlation between oral motor function and Aiming and Catching, $r(38) = .405$, $p = .012$, and a strong positive correlation with Balance, $r(37) = .616$, $p < .001$. No significant relation was observed between oral motor function and Manual Dexterity.

Relation Between Speech and Motor Abilities
Spearman’s rho correlations were conducted between speech and motor measures. Findings revealed significant moderate negative correlations between the number of CAS features and Manual Dexterity, $r(38) = -.4366$, $p = .024$; Aiming and Catching, $r(40) = -.564$, $p < .001$; and Balance, $r(40) = -.473$, $p = .002$. Likewise, GFTA-3 standard scores were moderately positively correlated with Manual Dexterity, $r(38) = .426$, $p = .008$; Aiming and Catching, $r(40) = .506$, $p = .001$; and Balance, $r(40) = .486$, $p = .001$. Finally, inconsistency severity percentages were also moderately negatively correlated with Manual Dexterity, $r(38) = -.366$, $p = .024$; Aiming and Catching, $r(40) = -.564$, $p < .001$; and Balance, $r(40) = -.473$, $p = .002$.

Relation Between Language Ability and Motor Performance
Spearman’s rho correlations were used to determine relations between language and motor abilities across groups. Results showed significant moderate-strong positive correlations between Expressive Language and Manual Dexterity, $r(35) = .655$, $p < .001$; Aiming and Catching, $r(37) = .487$, $p = .002$; and Balance, $r(37) = .672$, $p < .001$. Results also revealed moderate to strong positive correlations between
Receptive Language and Manual Dexterity, r(35) = .496, p = .002; Aiming and Catching, r(37) = .570 p < .001; and Balance, r(37) = .651, p < .001.

**Motor Performance in Children with CAS and SSD Reclassified on the Basis of Language Performance**
Participants with CAS and SSD were reclassified into groups on the basis of receptive language abilities; typically developing participants all evidenced typical language as required for assignment to the original TD group and therefore maintained their group assignment in the TD group. Kruskal-Wallis and Bonferroni-adjusted Mann-Whitney U tests were used to detect group differences in motor performance; the Bonferroni adjustment (.05/10 pairwise comparisons) resulted in setting a significance level of p = .005 for post hoc tests. Findings differed from those observed when children with normal and disordered language were grouped together. A group effect was detected for the Manual Dexterity (p = .010), Aiming and Catching (p = .007), and Balance (p = .001) components. As displayed in Figure 2, the CAS + LI group scored significantly lower than the TD group on all motor measures (p < .002). The CAS + LI group also scored lower than the SSD-only group on Manual Dexterity (p = .001) and Balance (p < .001), but not on Aiming and Catching (p = .007) once the Bonferroni correction was taken. Although the CAS + LI group, on average, tended to perform more poorly than the CAS-only group, no statistically significant differences between these groups were detected for any component. Likewise, no statistically significant differences were detected between the CAS + LI and SSD + LI groups or between the CAS-only, TD, SSD-only, and SSD + LI groups relative to each other.

All seven of the children in the CAS + LI group evidenced Movement ABC-2 total scores of 5 or below, indicating significant movement difficulty; in contrast, one of three children in the CAS-only group scored in this range. One of 10 children in the SSD-only group scored 5 or below compared to two of six children in the SSD + LI group. Even though there was not a significant statistical difference between the CAS and CAS + LI groups, it may be clinically meaningful that all seven children in the CAS + LI group scored in the "significant movement difficulty" range compared to one of three children in the CAS-only group.

**Discussion**
We posited that children with CAS would perform significantly more poorly than the TD and SSD groups on motor tasks and, in particular, on fine motor tasks (i.e., Manual Dexterity subtests). Data partially support our hypotheses. While the original CAS group did not perform significantly worse than the control groups on Manual Dexterity tasks (i.e., fine motor), they did evidence significantly poorer performance than other groups on the Balance (i.e., foundational skill required for gross motor competence) and Aiming and Catching (i.e., gross motor) components. In addition, on average, the CAS group scored more than 1 SD below the mean on all components of the Movement ABC-2, whereas the SSD and TD groups scored within the normal range on these motor assessments, indicating clinically meaningful differences between groups. When participants were reclassified on the basis of language ability, the CAS + LI group evidenced significantly poorer performance on the Manual Dexterity, Balance and Aiming, and Catching components compared to the TD group and demonstrated poorer performance on the Manual Dexterity and Balance tasks compared to children with SSD only; no statistically significant differences were observed for the CAS-only and SSD + LI groups relative to other groups for movement tasks. Although we may not have observed statistically
significant differences between some of our small groups, it is important to recognize that all seven children in the CAS + Li group evidenced motor impairments compared with only two of six children in the SSD + Li group, suggesting that language impairment alone did not likely account for the substantially higher percentage of children with motor impairments observed in the CAS + Li group. In addition, only one of three children in the CAS-only group scored in the test's "red zone" compared with all seven children in the CAS + Li group. Although only small samples were included in this study, this finding represents preliminary evidence that there may be a clinically meaningful difference in the motor abilities of subgroups of children with CAS (i.e., with and without language impairment) as children who score in the "red zone" are typically those who require intervention (Henderson et al., 2007, p. 99).

Receptive and Expressive Language abilities were each moderately-strongly correlated with all Movement ABC-2 components, wherein children with poorer language also evidenced poorer motor abilities and those with better language evidenced better motor abilities as well. Speech severity measures in terms of number of CAS features displayed and articulation test standard scores were also moderately correlated with all motor components of the Movement ABC-2. Finally, oral motor function was positively related to Aiming and Catching and Balance, but not Manual Dexterity.

The current results are consistent with extant research that shows poor motor abilities in children with CAS (Bradford & Dodd, 1996; Gretz, 2013; Potter et al., 2013) and represents new and important findings that complement previous work, which showed poorer motor skills in children with CAS, but not performance below the normal limit (Tükel et al., 2015). Likewise, findings partially support research that showed a positive relation between oral motor function and motor ability. Newmeyer et al. (2007) found that nonspeech oral sequencing was correlated with fine motor performance in children with severe speech disorders, whereas speech sequencing and fine motor performance were not related. The current work found significant correlations between speech ability and all Movement ABC components and that oral function was significantly positively correlated with Aiming and Catching and Balance, but not Manual Dexterity. Newmeyer et al. suggested that poor performance on oral sequencing and fine motor function tasks could relate to mirror neuron dysfunction that impacts imitation on these motor tasks; theoretically, this could potentially impact imitative speech tasks as well, although Newmeyer et al. did not find that poor speech sequencing was related to fine motor dysfunction in their study. Mirror neurons, located in the motor system, fire when executing and perceiving a motor act, even if the motor act has never been performed before (Newmeyer et al., 2007; Rizzolatti, Fabbri-Destro, & Cattaneo, 2009). Consequently, if there is a disruption to this system, it could negatively impact one's ability to correctly imitate motor tasks and speech models (e.g., those provided during treatment sessions). In the current study, children were provided with visual models for all Movement ABC-2 tasks and for oral function tasks as needed. Real words were elicited in a picture naming format (e.g., on the GFTA-3) or as auditory targets presented over the sound field (e.g., build upon words). If children with CAS experience difficulty with their mirror neuron system, it could result in poor motor performance across domains, as we observed in this study; alternatively, distinct factors and mechanisms could impact speech versus fine/gross/oral motor performance, as suggested by Newmeyer et al. That is, speech production may also activate language centers in the brain, whereas this would not be expected for fine/gross/nonspeech oral movements. Future research should investigate mirror neurons in children with CAS to determine the extent to which this system functions
properly during various speech and nonspeech motor tasks so that findings can be considered when developing targeted treatments for this population.

Previous findings on motor abilities in children with CAS are equivocal (Bradford & Dodd, 1996; Dewey et al., 1988; Gretz, 2013; Potter et al., 2013; Tükel et al., 2015). Differences between our findings and other studies may have been due to differences among testing instruments, diagnostic differences, or the symptom complexity of participants. Although they included children with oral motor sequencing issues, Newmeyer et al. (2007) did not differentially diagnose CAS from SSD; consequently, they may have investigated a different population than that which was examined in the current work. Alternatively, their use of the PDS, found to be less sensitive in identifying mild motor impairments compared with the Movement ABC (Van Waelvelde et al., 2007) employed in the current study, may have contributed to divergent findings. Tükel et al. (2015) restricted their sample to limit language impairment and exclude those who had previously undergone physical or occupational therapy and thereby may have limited the severity of their participants. The current study did not restrict participants based on language or therapy history, yet only three children had previously undergone physical or occupational therapy despite parents reporting concerns about their child’s motor abilities on our case history form. Future research should consider why children whose parents have concerns about motor ability do not receive motor evaluations. Do families report their concerns to pediatricians and evaluations are not deemed necessary? Do children meet certain motor milestones (e.g., crawling, taking steps) such that a referral to physical or occupational therapy is deemed unnecessary even though the quality of the child’s motor skills is poor? Are referrals made but, due to time constraints and other obligations, the evaluation is never completed? Our findings are consistent with previous work showing that pediatricians make more referrals to speech therapists compared with other allied health professions (Michaud & Committee on Children with Disabilities, 2004); consequently, speech pathologists have early and unique access to children who may be in need of physical and occupational therapy but whose pediatricians have not yet identified this need. Interprofessional practice, education, and collaboration can yield the highest quality care for our clients, but in order for us to collaborate to achieve optimal outcomes, we may first need to make a referral for a screening or assessment to a physical or occupational therapist. From there, we may be able to develop an integrated plan of care (ASHA, 2016).

Given that the current findings demonstrate a high prevalence of motor impairments in participants with CAS who have comorbid language impairment, it is likely that this represents the biggest difference between Tükel et al.’s (2015) participant sample and ours. The positive correlation we detected between motor and language abilities suggests that, by excluding children with the poorest language, we might also thereby exclude our poorest motor performers as well. We are confident in the internal validity of the motor assessment given that the Movement ABC requires demonstration for each task, a practice phase, and minimal verbal instruction and has previously been shown to be valid in children with low cognitive-linguistic abilities (Spand et al., 1999). The current findings provide further support for the use of the Movement ABC to detect motor impairments in children with a range of motor and learning abilities (Van Waalvelde et al., 2007) and suggest that physical and occupational therapy screenings should be considered for children with comorbid speech and language disorders and especially those with CAS + LI, if they are not already being seen by these allied health professionals.
Comorbid Speech, Language, and Motor Impairments in Children With CAS

We observed a high rate of comorbid speech, language, and motor impairments among our participants with CAS. It is well established that there is great interaction between the motor and cognitive-linguistic systems during development and that motor disorders are prevalent among children with language impairment (Hill, 2001; Powell & Bishop, 1992; Vuolo, Goffman, & Zelaznik, 2017). Previous studies have demonstrated poor manual dexterity (Owen & McKinlay, 1997; Powell & Bishop, 1992), balance, and gross motor ability in this population (Powell & Bishop, 1992). In fact, a recent study of 27 children with specific language impairment between 4 and 5 years of age showed that nine evidenced motor performance greater than 1 SD below the mean on the Movement ABC-2, indicating significant motor deficits in over 30% of participants with this disorder (Vuolo et al., 2017). These findings are consistent with the prevalence of disordered movement performance we observed in our SSD + LI group. In contrast, our data on children with CAS + LI showed even more pervasive and severe motor disturbances among 100% (7/7) of participants in this subgroup. Findings support the possibility of a third-factor, higher order deficit that mediates cognitive-linguistic and motor ability in children with CAS. Just as the procedural learning deficit hypothesis has been used to explain comorbid cognitive-linguistic and motor deficits in children with specific language impairment, developmental coordination disorder, and dyslexia (Hardiman, Hsu, & Bishop, 2013; Steinmetz & Rice, 2010; Ullman & Pierpont, 2005), it may also help to explain co-occurring speech, language, and motor impairments in children with CAS as well. Based on the current data, we would expect that children in the CAS + LI group would have poorer procedural learning ability than the CAS-only group. Ongoing work is testing this hypothesis in children with CAS with and without language impairment. Mirror neuron dysfunction—discussed above—does not preclude a procedural learning disturbance—rather, we posit that it would interact with and further compound procedural learning challenges in many contexts (e.g., treatment sessions, learning of motor skills).

It is notable that there were no females in the CAS-only group (n = 3) and that there were three in the CAS + LI group (n = 7). Although the samples are small, the discrepancy in the percentage of females between groups provides potential support for the female protective model (Jacquemont et al., 2014). This model proposes that a larger deleterious genetic mutation is required for females to express a disordered phenotype compared to the size of the mutation required for male peers to be symptomatic (Jacquemont et al., 2014). Consequently, there tend to be fewer females with certain diagnoses (e.g., autism), but when the mutation is large enough for the phenotype to be expressed, it results in a more severe expression of symptoms. In the current study, a larger genetic mutation may account for the greater overall complexity and severity of children in the CAS + LI group. Further scientific inquiry is required to determine the validity of this model in children with CAS.

Motor Tasks as Analogs to Speech Production

Children with CAS + LI scored poorly on all subtests of the Movement ABC-2. The Aiming and Catching and Balance components of this test assess body control and posture, limb function, spatial accuracy, control of force and effort, and timing of actions. Many of the skills probed in this assessment may be considered analogs for skills required for speech production. For instance, while vowels are often considered an easy type of speech sound mastered by the age of 3 years (Pollock & Berni, 2003), they require precisely graded movements of the tongue and jaw, without benefitting from physical contact.
with the teeth to guide tongue placement; aspects of this posturing and coordination may seem similar to how someone positions their hands for catching or holds and adjusts their body while performing a balancing task. Although there seem to be parallels between the speech and limb movements we observed, we recognize that motor control is task specific and that speech and limb movements rely on different structures and patterns of neural activation (Bunton, 2008; Grimme, Fuchs, Perrier, & Schöner, 2011; Newell, 1989; Ziegler, 2003). Even though these systems are distinct, this does not preclude the possibility of brain differences or a higher order deficit (e.g., procedural learning impairment) mediating motor and cognitive-linguistic learning across multiple systems (Leiner et al., 1991; Nicolson & Fawcett, 2007), which might explain the high rate of co-occurring fine/gross motor deficits observed in our participants with CAS + LI. Future research is needed to determine and better understand these possible mechanisms in children with CAS.

Limitations and Future Directions
The external validity of our current study is limited by small group size. It is important to replicate our findings in larger samples of children with CAS and SSD, with and without language impairment. It is also essential that we consider alternative methods for differential diagnosis of language impairment in children with severe speech disorders rather than rely on a solitary index measure, as we did in this study. Many of the diagnostic measures that incorporate language samples or story retell tasks can be difficult to score in children with severe speech disorders, but efforts should be made to incorporate more dynamic and comprehensive measures if possible.

Future studies in collaboration with physical and occupational therapists should carefully characterize the fine/gross motor responses evidenced by children with CAS and consider them in relation to perceptual and acoustic speech output. Likewise, research should aim to better understand the functional limitations of motor impairments among children with CAS (e.g., does gesture and/or sign usage differ between children with better/worse motor ability). Based on our data and given the high percentage of parents who reported motor concerns on their case history forms, it is possible that at least a subset of children with CAS experience developmental coordination disorder, which is characterized by motor performance that is 2 or more SDs below the mean and which leads to difficulty in completing activities of daily living or in the academic environment (American Psychiatric Association, 2013; Kirby & Sugden, 2007). By collaborating with physical and occupational therapists, we can better understand the motor profiles associated with this population and work together to provide optimal care for each individual.

Conclusion
The current study found compelling evidence of motor impairment among children with CAS + LI and, to a lesser degree, among small samples of children with CAS only and SSD + LI. Data support screenings or assessment of motor skills by physical and occupational therapists for these populations. While it is not within our scope of practice as speech pathologists to provide physical or occupational therapy, we can and should engage and refer to other health professionals, as needed, if these practitioners are not already part of the child's care team. Going forward, it is essential that we continue to learn about comorbid impairments in children with CAS and other SSD as these increase the likelihood of academic, social, emotional, and vocational challenges across the life span in these populations (Felsenfeld, Broen, & McGue, 1994; Lewis, Freebairn, & Taylor, 2000; Sices, Taylor,
Freebairn, Hansen, & Lewis, 2007). Likewise, future work should investigate whether a higher order third factor, such as poor procedural learning ability, may be negatively impacting learning across multiple modalities in children with CAS and other communication disorders.

Acknowledgments

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References


## Figures and Tables

### Table 1. Demographic and speech-language data by participant.

<table>
<thead>
<tr>
<th>Group</th>
<th>Subgroup</th>
<th>Age (months)</th>
<th>Sex</th>
<th>GFTA-3 SS</th>
<th>Average no. CAS features(^a)</th>
<th>Phonemic inconsistency %(^b)</th>
<th>Expressive Language SS(^c)</th>
<th>Receptive Language SS(^c)</th>
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<td>35.8</td>
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<td>19.5</td>
<td>45</td>
<td>55</td>
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<td>29.3</td>
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<td>50.0(^d)</td>
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<td>31.7</td>
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Note. GFTA-3 = Goldman-Fristoe Test of Articulation–Third Edition (Goldman & Fristoe, 2015); SS = standard score; CAS = childhood apraxia of speech; LI = language impairment; M = male; F = female; SSD = speech sound disorder; TD = typical development. a Number of CAS features was elicited in productions of the GFTA-3, customized lists of real words and nonwords (Nelson et al., 2016) that varied in complexity and number of syllables, and language sample based on Park Play picture description (Patel & Connaghan, 2014). b Phonemic inconsistency was determined using the inconsistency severity percentage, calculated on whole-word responses from the GFTA-3 (IuzziniSeigel et al., 2017). c Expressive and receptive language standard scores from the Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Wiig et al., 2004) or the Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013) d Participant 009 evidenced an average of 5.6 CAS features, including a severe prosodic disturbance. He had previously participated in therapy and had made substantial progress in articulation accuracy and evidenced low phonemic inconsistency (5.9%). To ensure appropriate group assignment, lexical inconsistency was evaluated across two productions of the multisyllabic word list. e Sum of Identification Core scores from the Test of Integrated Language and Literacy Skills (Nelson et al., 2016). Cut score to diagnose language/literacy disorders is 24 for children aged 72–95 months and 34 for children aged 96–143 months, indicating normal language for the three participants who underwent assessment with this testing instrument. f Participant had previously participated in therapy for SSD and exhibited poor intelligibility in connected speech, resulting in his assignment to the SSD group despite his GFTA-3 score of 95.

Table 2. Demographic and speech-language data by group.

<table>
<thead>
<tr>
<th>Variable</th>
<th>CAS (n = 10)</th>
<th>SSD (n = 16)</th>
<th>TD (n = 14)</th>
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<td>0.34 (0.8)</td>
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<td>Expressive Language SS</td>
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<td>111 (14)</td>
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<tr>
<td></td>
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<td>Group 2</td>
<td>Group 3</td>
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<tr>
<td>--------------------------</td>
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<td>---------</td>
<td>---------</td>
</tr>
<tr>
<td>Receptive Language SS</td>
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<td>96 (18)_{a,c}</td>
<td>114 (10)_{b,c}</td>
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<td>23 (1)</td>
<td>24 (0.5)</td>
</tr>
<tr>
<td>Oral Function score</td>
<td>27 (5)_{a,b}</td>
<td>31 (1)_{a}</td>
<td>32 (1)_{b}</td>
</tr>
</tbody>
</table>

Note. Group averages listed with standard deviations in parentheses. Groups sharing the same subscript letter were statistically different for the specified variable. Nonverbal IQ: from Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003); Articulation SS: from the Goldman-Fristoe Test of Articulation–Third Edition (Goldman & Fristoe, 2015); CAS Features: Iuzzini-Seigel et al. (2017); Inconsistency Severity %: Iuzzini and Forrest (2010); Expressive Language and Receptive Language SS: from the Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Wiig et al., 2004) or the Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013) for participants older than 6 years of age; Oral Mechanism Structure score: from Robbins and Klee (1987), the highest possible score is 24 and scores are expected to be 20–24 for this age range; Oral Function score: from Robbins and Klee (1987), the highest possible score is 32, and no age norms are available for this measure. CAS = childhood apraxia of speech; SSD = speech sound disorder; TD = typical development; SS = standard score; M = male; F = female
Figure 1. Movement Assessment Battery for Children–Second Edition (Movement ABC-2) component scores by group. The red line marks the cutoff for the “red zone” wherein scores of 5 or below indicate significant movement difficulty; a score of 6 indicates a high risk for movement difficulty and that performance should be monitored. Brackets indicate significant differences between groups. Error bars report standard error. CAS = childhood apraxia of speech; SSD = speech sound disorder; TD = typical development.
Figure 2. Movement Assessment Battery for Children–Second Edition (Movement ABC-2) component scores by groups reclassified based on language ability (CAS-only: n = 3; CAS + LI: n = 7; TD: n = 14; SSD-only: n = 10; SSD + LI: n = 6). The red line marks the cutoff for the “red zone” wherein scores of 5 or below indicate significant movement difficulty; a score of 6 indicates a high risk for movement difficulty and that performance should be monitored. Brackets indicate significant differences between groups. Error bars report standard error. CAS = childhood apraxia of speech; LI = language impairment; SSD = speech sound disorder; TD = typical development.