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Retrospective Case–Control Study of Communication and Motor Abilities in 143 Children With Suspected Childhood Apraxia of Speech: Effect of Concomitant Diagnosis

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Abstract

Purpose: This study sought to determine if children with childhood apraxia of speech (CAS) plus another major diagnosis (CAS+) are equivalent in communication and motor profiles to those with a primary diagnosis of CAS and no indication or report of any other diagnosis (CAS-Primary).

Method: This retrospective case–control study included a chart review of 143 children who were suspected of having CAS at Children's Hospital-Wisconsin between 1998 and 2004. Participants were between 30 and 127 months old and included 107 males. Participants were assigned to the suspected CAS-Primary group ($n = 114$) if they had characteristics of CAS but no other major diagnosis (e.g., galactosemia) and to the CAS+ group ($n = 29$) if a comorbid diagnosis was present. Groups were compared across demographic, communication, and motor characteristics.

Results: Children with CAS+ evidenced more severe motor profiles than those with CAS-Primary, $\chi^2 = (1, n = 122) = 4.952$, $p = .026$, and a small-to-medium effect size ($\Phi = .201$). On average, communication profiles also tended to be more severe among those with CAS+ wherein receptive language was poorer and phonemic inventories were smaller than those with CAS-Primary.

Conclusions: These retrospective data suggest that comorbid diagnosis may play an important role in communication and motor development in children with suspected CAS. These exploratory findings should motivate future prospective studies that consider the role of concomitant diagnoses in symptom profile and response to treatment in children with CAS.

Childhood apraxia of speech (CAS) is a speech motor planning disorder that can be associated with neurological injury/trauma, or complex genetic, metabolic, and

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Author Contributions

Jenya Iuzzini-Seigel: Formal analysis (Lead), Methodology (Lead), Writing – original draft (Lead). **Amy L. Delaney:** Conceptualization (Supporting), Investigation (Lead), Methodology (Equal), Project administration (Equal), Writing – review & editing (Equal). **Ray D. Kent:** Funding acquisition (Lead), Project administration (Supporting), Writing – review & editing (Lead).

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neurobehavioral disorders; the majority of cases, however, are idiopathic with no known cause (American Speech-Language-Hearing Association [ASHA], 2007). Although CAS affects approximately one child out of 1,000 (Shriberg et al., 2019), it occurs at a much higher prevalence when comorbid or co-occurring with certain diagnoses. For example, CAS has been observed in over 30% of children with Down syndrome (Wilson et al., 2019) and in up to 63% of children with galactosemia (Shriberg et al., 2011; Webb et al., 2003).

Children with CAS evidence a broad constellation of speech (ASHA, 2007; Murray et al., 2021), language (Lewis et al., 2004; Zuk et al., 2018), attention (Teverovsky et al., 2009), early feeding difficulties (Highman et al., 2008; Teverovsky et al., 2009), and generalized motor symptoms (Iuzzini-Seigel, 2019; Teverovsky et al., 2009; Tükel et al., 2015), as well as varied response to treatment (e.g., Iuzzini & Forrest, 2010; Maas & Farinella, 2012; Maas et al., 2014).

There are numerous features associated with CAS such as vowel errors, voicing errors, limited vowel and consonant inventories, delayed production of first words, reduced speech intelligibility, syllable segregation, high inconsistency, difficulty initiating an utterance or making coarticulatory transitions, groping, stress errors, and prosodic disturbance (e.g., ASHA, 2007; Highman et al., 2008; Iuzzini-Seigel & Murray, 2017; Murray et al., 2015, 2021; Overby et al., 2020). Expressive language disorder is present for the great majority of children with CAS, and research shows that standardized testing reveals receptive language disorder in a large subset of children with CAS as well, although receptive abilities are commonly better than expressive abilities (e.g., ASHA, 2007; Iuzzini-Seigel, 2021; Lewis et al., 2004; Zuk et al., 2018). Speech perception deficits are present in children with CAS and comorbid language disorder but appear to be less common in children with CAS with normal language performance (e.g., Zuk et al., 2018). Finally, recent research shows fine and gross motor deficits and an increased prevalence of developmental coordination disorder diagnosis among 50%–80% of children with CAS (Duchow et al., 2019; Iuzzini-Seigel, 2019). To date, it is unclear what drives divergent symptom profiles and varied response to treatment in this population, although recent work suggests that the Procedural Learning Deficit Hypothesis may apply (Iuzzini-Seigel, 2021).

Procedural learning is the process by which we implicitly acquire patterns through repeated practice (Nicolson & Fawcett, 2007) and is involved in the learning of grammatical rules, motor tasks (e.g., bike riding, hand games), and speech sound acquisition. Consequently, a procedural learning deficit would be expected to impact multiple systems, and the Procedural Learning Deficit Hypothesis has been used to explain co-occurring motor and cognitive-linguistic deficits in children with dyslexia, specific language impairment, and attention-deficit/hyperactivity disorder (ADHD; Nicolson & Fawcett, 2007) and most recently in children with CAS (Iuzzini-Seigel, 2021). Procedural learning deficits may also help to explain why children with CAS require such frequent and intense practice to learn speech targets during intervention (Iuzzini-Seigel, 2021; Maas et al., 2014).

Research on CAS has generally focused on determining core speech features of the disorder (Allison et al., 2020; Murray et al., 2021) and, in an effort to recruit larger participant samples, sometimes includes children with concomitant diagnoses (e.g., autism,

galactosemia). Consequently, it is unknown whether children with CAS plus another major diagnosis (CAS+) are equivalent in communication and motor profiles to those who have a diagnosis of CAS only (CAS-Primary). Some diagnoses are associated with complex challenges such as cognitive impairments and/or structural orofacial anomalies (e.g., Down syndrome), and therefore, prognosis and treatment may differ between children with and without concomitant diagnoses; other comorbid diagnoses (e.g., ADHD) may seem mild by comparison but still may impact treatment response. Although it may seem like common sense that children with CAS+ will evidence increased severity of communication and motor impairments and greater delays in meeting milestones compared to children with CAS-Primary, to date, these differences have not been reported in the data. It is essential that children with CAS-Primary and CAS+ are carefully studied and compared so that findings may inform development of targeted assessment protocols, treatment plans, and prognostic indices.

Developmental language disorder is found to occur among 50%–80% of children with CAS (ASHA, 2007; Iuzzini-Seigel, 2019, 2021; Lewis et al., 2004; Shriberg et al., 2019), compared to the already high rate of 1/13 in the general population of children (Tomblin et al., 1997). To date, it is unknown to what extent a co-occurring medical diagnosis may further increase the odds that a child with CAS will evidence language deficits as well. The effects of pervasive language disorder are serious and long term. Research shows that adults with a childhood history of developmental language disorder experience ongoing challenges including language impairments, literacy impairments, theory of mind deficits, worse social adaptation, lower rates of independent living, and more periods of prolonged unemployment compared to their siblings and controls with normal language abilities (Bishop & Adams, 1990; Clegg et al., 2005; Conti-Ramsden et al., 2018; Felsenfeld et al., 1992; McGregor, 2020; Whitehouse et al., 2009). Still other studies show an increased risk of psychiatric disorders (e.g., ADHD, social phobia) in children with a history of speech and language disorders (Snowling et al., 2006). Given that developmental language disorder is an extremely high prevalence disorder that has such wide-reaching and ongoing challenges associated with it, it is essential to determine whether this disorder co-occurs more commonly among children with CAS+ than CAS-Primary.

The current exploratory study aims to take a first step in empirically filling this gap by comparing the developmental communication and motor profiles of children with a diagnosis of suspected CAS-Primary to those with CAS+. If these groups vary in their involvement and severity, findings may support the development of distinct treatment approaches (e.g., Murray & Iuzzini-Seigel, 2017) and prognostic indices for subgroups of children with CAS, which are currently absent in the field. We posit that those with concomitant diagnoses will evidence greater severity of communication and motor impairments relative to those with CAS-Primary. The occurrence of CAS in different disorder groups may also hold clues regarding its nature and origin. See Supplemental Material S1 for brief capsule descriptions of diagnoses in which CAS has previously been reported.

Method

This study used a retrospective case–control design to investigate the speech, language, and generalized motor profiles of 143 children suspected of having CAS at Children's Hospital of Wisconsin-Milwaukee (CHW), over a 7-year period (1998–2004). All participants included in the study were seen as outpatients for initial evaluation, as a second opinion, as a re-evaluation, or as part of the Autism Behavior Communication Team. Exclusionary criteria included children younger than 30 months, children with an acquired apraxia diagnosis, and data from evaluations conducted while a child was admitted as an inpatient.

Procedures completed by the second author (A. D.) included a review of each child's medical chart including their speech/language evaluation report. The medical chart was also searched for medical diagnoses, birth and medical status, other developmental evaluation reports, and prior speech/language evaluation reports. The medical chart review was utilized to fill in gaps of information that may not have been reported in the speech/language evaluation report. These data were collected prior to the onset of electronic medical records and so the level of detail reported for each child varied. For example, gestational age may have been reported in weeks, as term delivery, or not at all.

The speech/language evaluation report typically included reasons for referral, summaries of (a) a previous medical chart review, (b) departmental case history form, and (c) caregiver interview. The departmental case history form was collected for initial evaluations only and included demographic information regarding the family and patient, birth, medical, and developmental information such as gestational age, onset of communication and motor milestones, and developmental therapy status. Absence of information in a child's chart likely reflected either (a) a lack of concern regarding a topic (e.g., clinicians may have only reported on ear infections if a child had a history of ear infections and did not include a statement saying "no history of ear infections" if there was not a concern) or (b) that a topic was not investigated (e.g., ear infections may not have been inquired about if the caregiver did not report a concern). Standard clinical practice was to gather the most useful information possible to contribute to a differential diagnosis, within the time constraints of one evaluation visit. Both informal and formal assessment procedures were attempted including speech and language testing, an oral peripheral examination, speech/language sample, stimulability testing, and screening for feeding, voice, resonance, and fluency. All procedures were approved by the CHW Institutional Review Board.

Participants

Initially, 516 children were identified using International Classification of Diseases, Ninth Revision billing codes for "apraxia of speech" and "other speech disturbance," which was the agreed upon code at CHW for children with *suspected* apraxia of speech. From this cohort, 210 charts were randomly selected to undergo detailed analysis and 143 met inclusionary/exclusionary criteria. Of these participants, 22 were undergoing initial evaluation, 79 were seeking a second opinion, one was a re-evaluation, and nine were participating in the Autism Behavior and Communication Team (ABC Team). In addition, one child was undergoing both an initial evaluation and assessment in the ABC Team, and 17 were seeking a second opinion and assessment in the ABC Team. Details

regarding evaluation status were unavailable for 14 participants. History of previous speech treatment was noted for 81 participants, whereas 19 participants reported no previous speech treatment. Information regarding treatment history was unavailable for 43 participants.

The CAS or suspected CAS diagnosis was made by 13 clinical speech-language pathologists based on criteria specified by Davis et al. (1998) and included inconsistent speech errors, prosodic disturbance, significant difficulty imitating words and phrases, limited consonant and vowel repertoire, vowel errors, and groping. CAS features were not routinely recorded in the charts, and no checklist was used to record the presence of CAS features in different speech corpi as is used in our prospective studies (Iuzzini-Seigel, 2019, 2021; Iuzzini-Seigel & Murray, 2017). As such, other than vowel and voicing errors, which were extracted from the phonetic inventories and are reported in Table 2, on occasion notes were made about CAS features for a subset of 24 participants. For the children in this subset, inconsistent errors were reported for 71%, and increased errors with increased utterance length or increased complexity were noted for 63%. It should be noted that a lack of reporting on these and other features does not necessarily mean that the child did not demonstrate this feature as it could also mean that the clinician simply did not make note of it in their report.

Of the 13 clinicians, four primary clinicians made 88% of the diagnoses including one who served on CHW's multidisciplinary evaluation team and conducted 40% of the evaluations. These four clinicians served mainly as diagnosticians at CHW, and as a result in total, they completed an estimated 13,000 pediatric speech and language evaluations over the 6-year time frame of the study. The children in this study represent the fraction of children who were diagnosed with CAS or suspected CAS opposed to the thousands of children diagnosed with other speech sound disorders (e.g., phonological disorders, articulation disorders). Because we are comparing two groups of children who evidenced features consistent with CAS at the time and who were diagnosed by the same small group of diagnostic experts during the same time frame and because the diagnostic features used largely overlap with criteria currently used to make the CAS diagnosis (e.g., ASHA, 2007; Iuzzini-Seigel, 2019, 2021; Iuzzini-Seigel et al., 2017; Murray et al., 2021; Shriberg et al., 2011), we believe our comparison of developmental profiles of children with suspected CAS with and without additional diagnoses is valid. This exploratory retrospective study does have some limitations related to its method, but this investigation on a large data set of children with suspected CAS contributes a substantial amount of novel and valuable information to the field.

Participants were assigned to the CAS+ group if they had a major concomitant diagnosis reported (e.g., autism, galactosemia), and to the CAS-Primary group if no other diagnosis was noted. Consequently, the CAS-Primary group included 114 children and the CAS+ group had 29. Table 1 includes a list of all concomitant diagnoses and the number of children who were affected by each.

The majority of caregivers reported on birth history ($n = 134$). In total, 90% of children in the CAS-Primary and 92% of children in CAS+ were reported to have achieved full-term gestation (CAS-Primary, $n = 108$: *Mdn* = 34 weeks gestation, range: 23–40 weeks; CAS+, $n = 26$: *Mdn* = 38 weeks gestation, range: 31–41 weeks). Birth weight was reported for

100 children, with children with CAS-Primary having an average birth weight of 7.26 lb (range: 2.12–10 lb) and those with CAS+ averaging 7.16 lb (range: 3.3–10.15 lb). Presence or absence of birth complications was reported for 103 children; 15% of children in the CAS-Primary group (n = 84) and 21% of children in the CAS+ group (n = 19) were reported to have birth complications.

All charts reported the results of formal and/or informal speech and language testing, and 86 caregivers provided a history of the onset/absence/and timeframe for a child's first words. Spontaneous speech samples were attempted for all participants in a play activity or in response to a conversational prompt. At minimum, any spontaneous productions, if present, were transcribed, and results from stimulability testing were reported if the child tolerated the testing. Phonetic consonant and vowel inventories were compiled for 103 participants based on the results of any and all speech produced during the evaluation (see Table 2). Due to the severity of the participants, standardized speech assessments were rarely able to be administered in accordance with standardized testing procedures, and therefore, these measures were not viewed as a clinical priority. For a small subset of participants, a standardized speech test such as the Clinical Assessment of Articulation and Phonology (Secord & Donohue, 2002) was administered to help in ascertaining the speech inventory, but scores were not calculated or reported for these administrations, which did not adhere to standard procedures.

If tolerated, standardized language tests were administered; this was dependent on the child's severity level with more severe talkers less likely to participate in standardized testing. Specifically, 134 participants were reported to have undergone language testing using one or more formal assessments which included the Expressive Vocabulary Test (Williams, 1997; n = 2), Peabody Picture Vocabulary Test–Third Edition (Dunn & Dunn, 1997; n = 10), Expressive One Word Picture Vocabulary Test (Gardner, 1979; n = 14), Clinical Evaluation of Language Fundamentals–Fourth Edition (Semel et al., 2003; n = 12), Preschool Language Scales–Third Edition (Zimmerman et al., 1992; n = 92), Test of Language Development–Third Edition (Hresko et al., 1999; n = 3), Test of Language Competence–Expanded Edition (Wiig & Secord, 1985; n = 2), and the Rosetti Infant–Toddler Language Scale (Rossetti, 1990; n = 9). Children were rated as having delayed language based on their age equivalent score or standard score (SS) on a standardized assessment.

Statistical Analyses

Nonparametric Mann–Whitney U tests for comparison of independent groups were used due to heterogeneity of variance and unequal group sizes. Chi-square tests were used to determine the relation between groups and nominal variables such as "receptive language delay" or "early feeding difficulty." Bonferroni corrections were used to maintain family-wise error rates.

Results

The CAS-Primary group (n = 114) ranged in age between 30 and 118 months (Mdn = 41 months; SD = 20) and included 88 males. Children in the CAS+ group (n = 29) ranged

in age between 30 and 127 months (*Mdn* = 55 months; *SD* = 28) and included 19 males. A chi-square test revealed no association between group and gender ($p = .143$). A Mann–Whitney *U* test revealed that the CAS+ group was significantly older than the CAS-Primary group ($p = .006$) at the time of their evaluation.

A subset of families reported on early feeding skills (CAS-Primary $n = 26$; CAS+ $n = 12$); of these, early feeding difficulty was noted for 38% of CAS-Primary and 75% of CAS+. A chi-square test revealed a significant association between group and early feeding difficulties, $\chi^2 = (1, n = 38) = 4.385$, $p = .036$, and the phi test for effect size revealed a moderate effect ($\Phi = .340$).

Results from an oral structure assessment were reported for 119 children (CAS-Primary $n = 94$; CAS+ $n = 25$). Normal oral structure was reported for 89% of children with CAS-Primary and 88% of children with CAS+. Results of an oral tone assessment were reported for 88 children (CAS-Primary $n = 74$; CAS+ = 14). A chi-square test was used to determine whether either group was more likely to demonstrate abnormal oral tone. The Fisher's exact test was selected due to a violation of statistical assumptions for the Pearson chi-square statistic. Results showed that 16% of children with CAS-Primary evidenced abnormal oral tone compared with 36% of children with CAS+, a nonsignificant relation ($p = .134$, Fisher's exact test).

For children in the CAS+ group, a range of diagnoses was reported (see Table 1). Seizures were noted in eight children, autism spectrum disorders were reported in six, and ADHD affected five; all other diagnoses reportedly affected only one to two children.

A chi-square test was used to determine whether either group was more likely to demonstrate delayed production of first words. Results showed that 66% of children with CAS-Primary evidenced delayed speech production compared with 80% of children with CAS+, $\chi^2 = (1, n = 115) = 1.546$, $p = .214$), a nonsignificant relation. Estimated intelligibility was compared between groups for a subset of 34 participants, for whom these data were available. Intelligibility estimates were based on all speech production efforts made by the child. Thirteen percent of children in the CAS-Primary subset ($n = 26$) were considered 75%–100% intelligible, whereas no children in the CAS+ subset ($n = 8$) were considered this highly intelligible. Similar percentages of children in the CAS-Primary (57%) and CAS+ (60%) groups were considered less than 50% intelligible, indicating a severe speech deficit for the majority of participants in each of these subgroups (Shriberg & Kwiatkowski, 1982). The remaining participants in both groups were considered to have a moderate-to-severe speech deficit. See Table 2 for communication findings by group.

Over 60% of children in the CAS-Primary group ($n = 79$) produced /b, d, m/ whereas only /m, d/ were produced by this percentage of children in the CAS+ group ($n = 24$). Between 40% and 60% of children in the CAS-Primary group produced /p, n/, whereas this percentage of children with CAS+ produced /b, p/. Between 25% and 39% of children with CAS-Primary produced /t, g, k, w/, whereas this percentage of children with CAS+ group produced /t, k, n, h/. Between 15% and 24% of children with CAS-Primary produced /ʃ, f, h, j/, whereas the same percentage of children with CAS+ produced only /s, j/. All

remaining sounds were produced by less than 15% of participants in each group. Vowel productions varied by group as well, with the CAS+ group producing fewer discrete vowels than the CAS-Primary group. Between 25% and 35% of participants with CAS-Primary produced /a, o, i, ^/; in contrast, this percentage of children in the CAS+ group produced only /o, ^/. Remaining vowels were produced by less than 25% of children in each group. It should be noted that typically developing children, by contrast, are expected to produce all nonrhotic vowels (vowels without r-coloring) with nearly 100% accuracy by the age of 3 years (Pollock & Berni, 2003).

The Preschool Language Scale: Third Edition was the language test that was administered most often, with 92 children (CAS-Primary $n = 74$; CAS+ $n = 18$) completing a standardized administration of this assessment. The mean receptive language SS for the CAS-Primary group was 83, and 81 for those with CAS+. The mean expressive language SS for the CAS-Primary group was 72, and 67 for the CAS+. The remaining language assessments were each administered to five or fewer children or were administered in combination with a second assessment, and consequently, it was unclear for some which test the standard language scores listed in the chart referred to. The chi-square test to determine a relation between group and receptive language delay was not statistically significant, but findings may suggest a clinically meaningful difference as receptive delays were noted for 60% of children with CAS-Primary and 77% of children with CAS+, $\chi^2 = (1, n = 123) = 2.593$, $p = .107$. The relation between group and expressive language was also not significant, $\chi^2 = (1, n = 120) = .592$, $p = .442$; on average, 91% of children with CAS-Primary evidenced expressive language delay compared with 96% of children with CAS+.

Evidence of motor delay ($n = 122$) was noted if parents reported any delay based on the age at which their child began sitting, crawling, or walking and was indicated in 27% of children with CAS-Primary and 50% of children with CAS+, resulting in a significant relation with group, $\chi^2 = (1, n = 122) = 4.952$, $p = .026$, and a small-to-medium effect size ($\Phi = .201$). Mann–Whitney U tests were used to determine group differences in age for when children first crawled, sat unassisted, or walked. A Bonferroni correction (.05/3 = .016) was used to preserve the family-wise error rate. Significant group differences were noted for the age at which children first sat ($p = .002$). On average, children with CAS-Primary sat at 6.5 months, crawled at 8.5 months, and walked at 13.5 months, whereas those with CAS+ sat at 10 months, crawled at 15 months, and walked at 24.5 months.

Discussion

This study found that children with CAS+ evidenced more severe communication and motor profiles than those with CAS-Primary. Although the concomitant diagnoses represented in this study were varied, eight children had histories of seizures and five had a diagnosis of ADHD, consistent with previous literature showing neurological conditions and attentional deficits in this population (Teverovsky et al., 2009). Likewise, six out of 143 children had a reported diagnosis of autism spectrum disorders, which is over twice the rate that autism is found in the general population (Maenner et al., 2021). This finding provides further support for the literature showing a high rate of comorbid CAS in families with heritable genetic

diagnoses associated with autism (Boyar et al., 2001) but is in contrast to other research that shows no co-occurrence between CAS and autism (Shriberg et al., 2011, 2019).

A higher percentage of children in the CAS+ group (75%) experienced early feeding difficulties compared to those in the CAS-Primary group (38%), although this was not statistically significant. Feeding difficulties are often reported in the literature surrounding children with CAS (Forrest, 2003; Highman et al., 2008; Rosenbek & Wertz, 1972) and perhaps more commonly among children with oral apraxia (Arvedson, 2000; Wilson & Hustad, 2009). These findings align with Highman and colleagues' (Highman et al., 2008) report of parent questionnaire responses in which 55% of children with CAS-Primary demonstrated early feeding difficulties, as did 55% of children with specific language impairment, showing that feeding issues are not pathognomonic for children with CAS. Other research (Forrest, 2003) reports that some speech pathologists have considered discoordination during feeding as a differential diagnostic feature of CAS, although this is not common. The higher rate of early feeding difficulties in our CAS+ group can perhaps be used as a signal that children with this history are at greater risk for multisystem deficits rather than children with CAS as a primary or sole diagnosis and should be monitored and screened for motor and other impairments accordingly.

Data partially support our hypotheses that children with CAS+ have more severe communication profiles compared to those with CAS-Primary. Although the children in the CAS+ group were significantly older, they tended to have lower intelligibility and smaller vowel and consonant inventories compared to children with CAS-Primary. Difficulty producing the voicing distinction, another common feature of CAS was also frequently observed with /b, d, m/ being the only consonants used by > 60% of participants. Although intelligibility may correlate with inventory size or percent consonants correct, it is also likely to be impacted by other factors among children with CAS such as phonemic inconsistency, syllable segregation, and even language ability. Given the small number of children for whom intelligibility data were available in this study, we were not able to conduct additional analyses to determine what was driving the low intelligibility demonstrated by our participants; these are important variables to consider in future prospective research on children with CAS.

A higher percentage of children in the CAS+ group (80%) evidenced delayed production of first words compared to the CAS-Primary group (60%). While expressive language delays affected both groups at roughly the same rate (> 90%), receptive language delays appeared more prevalent in the CAS+ group compared to those with CAS-Primary; although this difference was not statistically significant, it may represent a clinically meaningful finding that will help us to create targeted prognostic indices and interventions for these populations in the future. In addition, low receptive language performance is often an exclusionary criterion for studies of CAS, and consequently, such studies may be excluding children who are very much representative of the CAS population, thereby limiting external validity of study findings. Future studies should consider relaxing this criterion to increase generalization.

Our findings are also consistent with the extant literature (Lewis et al., 2004; Morgan & Webster, 2018; Snowling et al., 2006) that shows a high rate of attention deficits among individuals with CAS. These results support the importance of ongoing educational and social service provision for individuals in this population to promote as much academic, social, and vocational success as possible (Clegg et al., 2005).

The CAS+ group was reported to have significantly worse fine and gross motor abilities than children with CAS-Primary such that children with comorbid impairments took nearly twice as long to begin to crawl and walk compared to children with CAS-Primary. Motor delays were noted in 50% of children with CAS+ and 27% of children with CAS-Primary, data that are consistent with some previous reports of motor deficits in children with CAS (e.g., Teverovsky et al., 2009; Tükel et al., 2015) and lower than others (e.g., Iuzzini-Seigel, 2019). Previous research (Iuzzini-Seigel, 2019) showed that underdiagnosis of motor deficits is a substantial issue among children with CAS with upwards of two thirds of participants with CAS scoring in the disordered range on a standardized motor assessment, even though they had not previously undergone a physical or occupational therapy evaluation or treatment. Consequently, this study may underreport motor impairments in our participants with CAS-Primary and CAS+ because such diagnoses have not yet been made. The extant literature notes that pediatricians are more likely to refer children to speech pathologists than to any other type of allied health professional (Michaud & Committee on Children With Disabilities, 2004). Consequently, it is possible that motor impairments are even more prevalent than the numbers we report because children had not been referred for physical or occupational therapy evaluations.

It is notable that the CAS+ group reported motor impairments at nearly twice the rate of those with CAS-Primary. This may be useful information to consider when developing prognostic indices and targeted assessments and treatments for these subgroups. In addition, several of the comorbid diagnoses reported in this study are independently associated with motor impairments. For instance, previous work (Vicari, 2006) shows that children with Down syndrome sit between 8 and 11 months, crawl between 12 and 17 months, and walk between 15 and 74 months. These children also often demonstrate hypotonia, which is known to impact achievement of motor milestones. Mild-to-severe motor impairments are also reported among half of children with ADHD, although medication has been found to improve motor skills in 50% of those affected (Kaiser et al., 2015). Research on outcomes of children with a history of motor deficits (with and without comorbid medical diagnoses) shows increased risk of academic difficulty, attention deficits, behavior problems, and social issues in this population (Dewey et al., 2002). Taken together, motor impairments—like speech and language impairments—substantially increase the risk of academic, social, vocational, and emotional challenges in adolescence and adulthood, and consequently, referrals to occupational and physical therapists should be made if concerns are noted. The consequences of fine motor deficits extend far beyond difficulty threading beads on a string or tying one’s shoelaces. If a child has poor manual dexterity, they may have poor control over handwriting, which can negatively impact their ability to take notes quickly and sufficiently during classes, limiting access to the curriculum. If handwriting is poor, they may have difficulty neatly lining up numbers, which will make math problems more onerous to complete and more susceptible to errors. Similarly, children with poor gross

motor skills are also prone to poorer fitness levels, potentially leading to lifelong health disparities, increasing risk for diabetes, stroke, and other health issues (e.g., Cantell et al., 1994; Haga, 2008; Kraus et al., 2019).

Although it is unknown what ties together speech, language, and motor deficits in children with CAS, recent research provides preliminary support for the Procedural Learning Deficit Hypothesis in this population (Iuzzini-Seigel, 2021). Consequently, it is critical to assess the full breadth of motor and cognitive-linguistic skills in this population, keeping in mind that motor and language disorders are undiagnosed in ~50% of children (Iuzzini-Seigel, 2019; Michaud & Committee on Children With Disabilities, 2004; Prelock et al., 2008; Tomblin et al., 1997). As speech pathologists are often the first referral made by pediatricians, it is essential that we make referrals to other allied health professionals if concerns are observed and that we consider potential pattern learning difficulties when designing treatment plans and scheduling treatment sessions. There is mounting research to support frequent and intense sessions that provide copious opportunities for practice for children with CAS to facilitate learning and generalization as these allow the increased exposure/practice to treatment targets that is needed to demonstrate and retain learning (Iuzzini-Seigel, 2021; Maas et al., 2014). Based on the extant research and the higher prevalence of multisystem differences found among children with CAS+ in this study, procedural learning may be poorer among children with CAS+ compared to CAS-primary. Future research should compare procedural learning abilities and treatment outcomes in children with CAS+ and CAS-primary.

It is notable that the CAS+ group had a lower ratio of males to females (2:1) compared to the CAS-Primary group (3:1). This discrepancy suggests consideration of the female protective model, wherein a larger genetic mutation is needed for females to express a disordered phenotype relative to the size of mutation that is required for males to be symptomatic (Jacquemont et al., 2014). For instance, a study of individuals with autism revealed that females evidenced a two- to threefold increase in large neurodevelopmentally deleterious copy number variants compared to males (Jacquemont et al., 2014); consequently, there are fewer females with autism, but those who are affected tend to be more severe than males with this diagnosis. The current data are consistent with the female protective model in that the CAS+ group demonstrated more severe communication and motor impairments and had a higher percentage of females than the CAS-Primary group.

Differential diagnosis of CAS from other speech sound disorders can be challenging in that many of the features that contribute to the diagnosis are not pathognomonic, occurring only among children with CAS. In the CAS+ population, differential diagnosis may be even more difficult because of potentially co-occurring dysarthria, orofacial dysmorphology, and even dysfluency. Are there any concessions that should be made when differentially diagnosing CAS in children with complex presentations compared to those who present with CAS-Primary? We would argue that the CAS profile of features (e.g., ASHA, 2007; Iuzzini-Seigel, 2019; Iuzzini-Seigel & Murray, 2017) should be apparent for any preschool-age or school-age child who warrants the CAS diagnosis. A child may evidence additional symptoms/deficits (e.g., dysarthria, dysfluency), but still, differentiating CAS features should be present regardless of comorbid diagnosis.

The current work has limitations related to the retrospective design. Whereas our prospective studies use a highly transparent, replicable, and detailed differential diagnostic procedure with high reliability (Iuzzini-Seigel, 2019, 2021; Iuzzini-Seigel et al., 2017), group assignment for participants in this study was based on Davis et al.'s features as determined by expert opinion. We lacked specific details as to which diagnostic criteria were demonstrated by each participant as these were not noted in each chart. We believe that this limitation was equal for both groups and, consequently, was unlikely to bias results. An additional limitation was the small sample size for some of our group comparisons that may have resulted in our failure to detect group differences due to low power.

In conclusion, this exploratory research shows that children with CAS+ tend to evidence more severe communication and motor profiles compared to those with CAS without another major diagnosis. This provides further motivation for consideration of subgroups in future studies that examine the nature and treatment of CAS and development of prognostic indices for this population. The research also revealed a high percentage of children with seizure disorders, autism spectrum disorders, and ADHD in the CAS+ group, although this group was small ($n = 29$). Going forward, it will be essential to conduct prospective studies that carefully phenotype the speech, language, and motor characteristics of children in these specific populations. In addition, it would be beneficial to determine the relation among procedural learning, communication, motor skills, and treatment outcomes for children with CAS-Primary and CAS+. Diagnosing and treating clinicians should consider care of the whole child when working with children with CAS and ensure that appropriate referrals are made to allied health professionals when indicated. For children who report early feeding difficulties, more severe speech and language history, delayed meeting of motor milestones, or presence of other diagnoses (e.g., seizure disorders), referrals to our nonspeech colleagues should be among our first considerations. Future prospective work is needed to determine differences in therapeutic outcomes and academic, social–emotional, and vocational success for children with CAS-primary versus those with CAS+.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Table 1.

Listing of the concomitant diagnoses evidenced by the CAS+ group and the number of children with each diagnosis.

Diagnosis	Children (*n*) for whom this diagnosis was primary diagnosis other than CAS	Children (*n*) for whom this diagnosis co-occurred with another major diagnosis[a]
ADHD	3	2
Learning disability	—	1
Seizures	4	4
ASD or PDD	1	5
Cerebral palsy	—	1
Cocaine exposure	—	1
Lead exposure	—	1
Unspecified chromosomal disease	1	—
Closed head injury	1	—
Congenital heart defect	—	1
Down syndrome	1	1
Dysphasia	—	1
Galactosemia	1	
Hypotonia	1	1
Mental retardation	—	1
Neurofibromatosis	1	—
Hearing loss	1	—
Hydrocephaly	1	—
Microcephaly	1	—
Leukemia	1	—
Motor apraxia	—	1
Left-sided paresis/hemiparesis	—	2
Intrauterine CVA	—	1

Note. CAS+ = childhood apraxia of speech plus another major diagnosis; CAS = childhood apraxia of speech; ADHD = attention-deficit/hyperactivity disorder; ASD = autism spectrum disorder; PDD = pervasive developmental disorder; CVA = cerebrovascular accident.

[a]Note that multiple diagnoses were reported for numerous children, and therefore, the sum of individual diagnoses exceeds the number of children in the CAS+ group.

Table 2.

Communication data by group.

Variable	CAS-Primary (*n*)	CAS+ (*n*)
Intelligibility estimate	**(*n* = 26)**	**(*n* = 8)**
	Percentage of children who demonstrated indicated level of intelligibility	
1%–24%	27%	10%
25%–49%	23%	38%
50%–74%	35%	50%
75%–100%	15%	–
Consonant production	**(*n* = 79)**	**(*n* = 24)**
Consonant produced by > 60% of participants	d, b, m	d, m
Consonant produced by 40%–60%	p, n	b, p
Consonant produced by 25%–39%	t, g, k, w	t, k, h, n
Consonant produced by 15%–24%	ʃ, s, f, h, j	s, j
Vowel production	**(*n* = 79)**	**(*n* = 24)**
Vowel produced by 25%–35% of participants	a, o, i, ^	o, ^
Vowel produced by 10%–24% of participants	e, u	a, i
Type of language delay [a]	**(*n* = 98)**	**(*n* = 26)**
Receptive language delay	60%	77%
Expressive language delay	91%	96%

Note. CAS-Primary = childhood apraxia of speech and no indication or report of any other diagnosis; CAS+ = childhood apraxia of speech plus another major diagnosis.

[a]Language delay based on standardized score or age equivalent from formal testing.