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## **Procedural Learning, Grammar, and Motor Skills in Children With Childhood Apraxia of Speech, Speech Sound Disorder, and Typically Developing Speech**

Jenya Iuzzini-Seigel

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## Research Article

# Procedural Learning, Grammar, and Motor Skills in Children With Childhood Apraxia of Speech, Speech Sound Disorder, and Typically Developing Speech

Jenya Iuzzini-Seigel<sup>a</sup> 

**Purpose:** This case-control study sought to determine if (a) children with childhood apraxia of speech (CAS), other speech sound disorders (SSDs), and typical development would perform differently on a procedural learning assessment and (b) whether grammatical ability would impact group differences.

**Method:** Communication, motor, and procedural learning abilities were assessed in 48 children with CAS ( $n = 13$ ), SSD ( $n = 20$ ), and typical development ( $n = 15$ ), between 43 and 97 months of age ( $M = 66$  months,  $SD = 12$  months).

**Results:** On average, children with CAS demonstrated grammatical and motor impairments and required an increased number of exposures to the visuospatial sequence to demonstrate procedural learning, compared to peers with SSD or typical development. A subset of children from each group demonstrated an unanticipated procedural learning

pattern wherein they evidenced an uptick in reaction time during the second sequenced block. Children with CAS with this pattern still evidenced procedural learning gains by the fifth sequenced block. In contrast, children with SSD and typical development with this pattern showed poor procedural learning outcomes and were characterized by lower scores on language and motor assessments as well.

**Conclusions:** This research provides partial support for the procedural learning deficit hypothesis in children with CAS and for a subset of children with SSD as well. Future research should examine the role of a serial reaction time task in identifying children at risk of multisystem communication and motor deficits.

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Childhood apraxia of speech (CAS) is a neurological speech disorder in which the motor planning and programming of speech movements is impaired without neuromuscular deficits such as abnormal tone (American Speech-Language-Hearing Association [ASHA], 2007). CAS manifests in a variety of symptoms such as inconsistent speech production, prosodic disturbance, and difficulty transitioning between sounds and syllables (ASHA, 2007). In addition, comorbid impairments such as language (Iuzzini-Seigel, 2019; Iuzzini-Seigel et al., 2017; Murray et al., 2019; Zuk et al., 2018), literacy (Miller et al., 2019), and fine/gross motor (Duchow et al., 2019; Iuzzini-Seigel,

2019; Teverovsky et al., 2009) deficits are observed in ~50% of children in this population. While typically developing (TD) children are able to acquire speech sounds, grammar, and motor patterns without being explicitly taught, children with CAS tend to require copious amounts of intensive treatment to make even minimal gains (Case & Grigos, 2016; Edeal & Gildersleeve-Neumann, 2011; Forrest & Iuzzini, 2008; Grigos & Kolenda, 2010; Iuzzini & Forrest, 2010; Maas et al., 2014; Murray et al., 2015; Strand & Debertine, 2000; Strand et al., 2006; Thomas et al., 2014). Some children in this population may show limited or no generalization (e.g., Ballard et al., 2010; Strand & Debertine, 2000) of treatment targets to untreated words, contexts (e.g., in a sentence or in conversation), and environments (e.g., outside of the treatment session), while other studies report generalization to untreated items in 25%–40% of participants depending on treatment type and time point when maintenance data were collected (Ballard et al., 2010; Maas & Farinella, 2012; McCabe et al., 2014; Murray et al., 2014; Preston et al., 2013; van Rees et al., 2012). Other studies,

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including the randomized control trial that compared Nuffield and ReST treatment for children with CAS showed accuracy gains of 15%–30% on real-word and pseudoword generalization probe items at 1 month posttreatment (Murray et al., 2015). The treatment intensity required and often slow gains can make treatment a long and arduous process requiring persistence, resilience, and ample investment of time and money by children/families, schools, and insurance companies. The mechanism that ties together these co-occurring speech, motor, and cognitive–linguistic impairments is unknown.

One possible mechanism linking together these seemingly divergent skills could be a deficit in procedural learning ability. Procedural learning is the system by which a variety of cognitive–linguistic and motor skills are implicitly learned and automatically produced after repeated exposure and practice (Sanjeevan & Mainela-Arnold, 2017; Ullman & Pierpont, 2005). The procedural learning system is implicated when we accurately type on a keyboard without looking at the keys, when we ride a bike, or when we play hand games such as “boom snap clap” or “Miss Mary Mack.” Likewise, the procedural learning system is used to acquire grammatical rules and speech sounds as well. The procedural learning deficit hypothesis suggests that a procedural learning impairment underlies multisystem motor and cognitive–linguistic deficits experienced by a variety of populations and has been used as one possible explanation for the co-occurrence of these deficits in children with developmental language disorder (DLD) and dyslexia (Nicolson & Fawcett, 2007; Sanjeevan & Mainela-Arnold, 2017). In this study, we test the procedural learning deficit hypothesis in children with CAS, in comparison to their peers with typical development and other speech sound disorders (SSDs).

### ***Procedural Learning in Children With DLD: Equivocal Findings***

Language deficits are reported in ~50% of children with CAS (ASHA, 2007; Murray et al., 2019), with some studies reporting even higher rates of this co-occurring disorder (Iuzzini-Seigel, 2019; Lewis et al., 2004). It is therefore essential that we understand the role of procedural learning in children with language deficits. Results vary for the extant research on procedural learning among children with DLD.<sup>1</sup> Equivocal procedural learning findings in

<sup>1</sup>DLD refers to language impairments that affect communication or learning in daily life, that are not associated with a broader developmental disorder (e.g., autism spectrum disorder), and that are unlikely to remediate without intervention (Bishop et al., 2017). DLD can co-occur with other issues such as speech deficits, attentional deficits, motor problems including developmental coordination disorder, reading and spelling problems, and/or behavioral and social–emotional issues. This term has gained favor relative to the term *specific language impairment* (SLI) as SLI was more exclusive and did not include children with co-occurring conditions or below-average IQ. The current paper will use the term *DLD* throughout, including in our discussion of the extant literature that may have used the terms *language impairment* or *SLI* to describe participants.

children with DLD are in large part due to differences in methods used to assess the procedural learning construct or in how groups were assigned (Hedenius et al., 2011; Hsu & Bishop, 2014; Sanjeevan & Mainela-Arnold, 2017; Ullman & Pierpont, 2005). Simple hand–eye coordination tasks often fail to find differences between children with DLD and those with typical development. For instance, Zelaznik and Goffman (2010) found that children with DLD perform comparably to TD peers in tapping and rhythmic circle drawing tasks. Similarly, children with DLD performed equivalent to TD peers on the pursuit rotor task (Hsu & Bishop, 2014). In this task, the participant uses a stylus pen to maintain contact with a dot that moves on a computer screen and requires the individual to adapt the arm’s rotational movement and speed to maintain that contact and ensure accuracy. Likewise, Sanjeevan and Mainela-Arnold (2017) assessed visual–motor adaptation ability using a mirror drawing task in which the participant was asked to draw two 4-pointed stars using only a mirror reflection. Findings revealed that children with DLD performed equivalent to TD peers on this task.

In contrast to performance on the simple tasks, more complex tasks have revealed group differences between children with DLD and those with typical development. For instance, in a novel knot tying task, children with DLD demonstrated lower accuracy than TD peers (Sanjeevan & Mainela-Arnold, 2017). Sanjeevan and Mainela-Arnold deduced that procedural learning impairments in children with DLD are restricted to complex sequence learning tasks and spare the visual–motor adaptation system.

The serial reaction time task (serial reaction time; Nissen & Bullemer, 1987) has long been considered the “gold standard” for assessment of procedural learning, but results have been equivocal in children with DLD. In this task, four squares are presented on a computer screen and a visual stimulus appears in one of the squares. The participant then uses a button response box to press the button that corresponds to the orientation of the stimulus on the screen, as quickly as possible. Following the participant’s response, the stimulus moves to a different position in either a sequenced or random order. This task continues for multiple blocks that each contains approximately 100 trials. A random block is introduced as the ultimate or penultimate block with the expectation that, if procedural learning has occurred, the reaction time of the sequenced blocks will get faster and faster with practice, but then there will be a rebound effect during the random block during which reaction time will increase again. Findings from the serial reaction time task tend to show that children with DLD require more exposure to the sequence to evidence learning (e.g., Gabriel et al., 2013; Hsu & Bishop, 2014; Tomblin et al., 2007), although some studies show similar performance to TD peers (Hedenius et al., 2011). Hedenius et al. initially found that children with DLD performed comparably well to TD peers on retention of a learned sequence. To further elucidate the extant equivocal findings of procedural learning studies in children with DLD, Hedenius et al. (2011) then recategorized participants on the basis of *grammatical* deficits. Results showed that those with TD grammatical ability evidenced

good procedural learning of a sequence during the initial experimental session and effective consolidation and sequence retention during a later session. In contrast, those with grammatical deficits evidenced poor procedural learning ability, and although they showed sequence learning during the first experimental session, they showed poor consolidation and retention at a subsequent session. In fact, after only 3 days, this subgroup failed to retain the sequence learned during the initial session.

Hedenius et al.'s findings are compelling in consideration of treatment outcomes in children with CAS who can be poor responders in therapy and who frequently show within-session performance gains but may fail to show longer term learning and generalization (Maas & Farinella, 2012; Preston et al., 2013). Children with speech disorders may receive treatment only 1–2 times per week, such that there can be a gap of several days or even a week between sessions. Even if a child starts to make gains within a treatment session, the gap between sessions could potentially prevent the child from retaining the newly learned information. Procedural learning findings may help to explain why some children with CAS show a greater benefit from shorter, more frequent therapy sessions relative to longer, less frequent sessions (Edeal & Gildersleeve-Neumann, 2011).

### ***Procedural Learning in Children With Motor Impairments: Equivocal Findings***

A small body of literature has investigated procedural learning ability in children with motor impairments such as cerebral palsy (CP; e.g., Gofer-Levi et al., 2013) and developmental coordination disorder (DCD; e.g., Wilson et al., 2003) and found equivocal results. Children with DCD, like those with CAS, typically make up a heterogeneous population wherein fine and/or gross motor abilities are impaired to the level that activities of daily living are affected. Anecdotal evidence reports that children with DCD demonstrate difficulty automatizing certain activities of daily living such as tying shoe laces, and neuroimaging studies have shown less activation of cerebellar-parietal and cerebellar-prefrontal circuits in this population—areas associated with visual-spatial learning (Zwicker et al., 2011).

Wilson et al. (2003) employed the serial reaction time task to investigate procedural learning ability in school-aged children (8–12 years old) with DCD. Findings showed procedural learning patterns that were comparable to TD peers, although reaction time per trial tended to be slower among children with DCD. In addition, Wilson et al. failed to introduce a random block toward the end of the task, and consequently, they were not able to confirm that implicit *sequence* learning occurred rather than a practice effect in which performance got faster on the task in general. Wilson et al. suggested that their findings supported appropriate functioning of the basal ganglia in their participants with DCD during this simple procedural learning task but indicated it was unknown whether this would be preserved during more complex tasks as well. Lejeune et al. (2013) also used the serial reaction time task to test procedural

learning in children with DCD, adapting the task to use a touch screen to record responses rather than the standard button response box, which requires greater perceptuomotor coordination and perhaps greater working memory as well (Gofer-Levi et al., 2013). Consistent with Wilson et al., Lejeune et al. also found that procedural learning appeared to be intact in their participants with DCD.

Examination of procedural learning in children with CP shows a different result (Gofer-Levi et al., 2013). Gofer-Levi et al. administered the serial reaction time task to 22 children and adolescents with spastic CP (ages 9–20 years) and 23 TD controls (ages 9–18 years). Findings showed that participants with CP had slower reaction time than controls and that while they did get faster at the task, they did not demonstrate the rebound effect (up-tick in reaction time) during the introduction of a novel sequence between the final two sequenced blocks. The authors instead attributed the performance gains in their CP group to explicit learning of instructions and integration of feedback but reported that this population “may not be sensitive to the ‘hidden’ sequence underlying common everyday procedures. Consequently, the order of things should be explicitly presented to them, step by step, to promote motor skill acquisition” (Gofer-Levi et al., 2013, p. 3677).

### ***Procedural Learning Impairment in Children With Comorbid Deficits***

The interaction between the linguistic and motor systems is well established (Iuzzini-Seigel et al., 2015; Nip et al., 2011; Smith & Goffman, 2004; Walsh et al., 2006; Zelaznik & Goffman, 2010; Zuk et al., 2018) and has been demonstrated through a variety of levels of analysis. Kinematic analysis of children with language deficits reveals evidence of decreased oromotor coordination (Alcock et al., 2000; Goffman, 1999; Zelaznik & Goffman, 2010), providing support for the high interconnectivity of the neural substrates of both systems (Arbib, 2006; Ojemann, 1984). Likewise, behavioral assessments of motor skills reveal poorer fine and gross motor performance among children with language disorder relative to TD peers (Iuzzini-Seigel, 2019; Powell & Bishop, 1992; Zelaznik & Goffman, 2010).

Ojemann (1984) posited that sequential movement and language share a common substrate in the lateral perisylvian cortex of the dominant hemisphere; consequently, a disturbance or underdevelopment of this region could yield co-occurring motor, speech, and language deficits. Nicolson and Fawcett (2007) developed a neural systems framework that links procedural learning deficits to the multisystem deficit profile commonly observed in children with dyslexia, DLD, and DCD. In this framework, there are two major routes posited for each of the corticostriatal and corticocerebellar circuits: a cognitive-linguistic route and a motor route (Balsters et al., 2010; Ramnani, 2006). Disorders may be characterized by a primary impairment of one route and a secondary impairment of the other. Nicolson

and Fawcett suggest that dyslexia is linked to a primary disturbance of a cognitive–linguistic route and that those with concomitant motor deficits have a secondary impairment of a motor route. One unexplored but compelling possibility is that the speech and fine/gross motor deficits (when present) in children with CAS are both associated with primary impairment to a motor route, and those with concomitant language deficits have a secondary impairment to a cognitive–linguistic route as well.

Peter and colleagues (Peter et al., 2018) found evidence of a global sequencing processing deficit in individuals with CAS; this central deficit is thought to underlie performance in a breadth of cognitive–linguistic and motor modalities such as reading, spelling, nonword repetition, and alternating syllable repetition. Peter and colleagues suggested that, because the sequential processing deficit was evident in a variety of modalities, the cerebellum—a key region that subserves movement and cognitive–linguistic performance (and the procedural learning system)—could be implicated (Leiner et al., 1991; Molinari et al., 1997).

### ***Evidence to Support a Procedural Learning Impairment in Children With CAS***

It is well established that acquisition and automaticity of motor sequences rely on procedural learning (Ullman & Pierpont, 2005). Unfortunately, children with CAS seem to lack speech automaticity as reflected by high speech inconsistency (e.g., inconsistent productions of the same target phoneme, word, or phrase; syllable segregation; and lengthened coarticulatory transitions; ASHA, 2007; Iuzzini-Seigel et al., 2017; Maassen et al., 2001). Terband et al. (2009) used the Directions into Velocities of Articulators model (Guenther, 2006) to explain speech deficits associated with CAS and attributed CAS symptoms to poor feedforward control resultant from weak sensory-motor projections. In a TD talker, sensory-motor neural traces are posited to strengthen over repeated productions as the child maps his/her productions onto corresponding adult forms (Guenther, 2006; Tourville & Guenther, 2011). Children then establish phonemic–articulatory mappings to produce meaningful and consistent phonemic contrasts. Both of these stages rely on procedural learning and yield increased efficiency/automaticity of speech as neural maps are strengthened. As such, a procedural learning deficit would be expected to negatively impact the ability to establish consistent and meaningful mappings, which could result in speech inconsistency—a hallmark feature of CAS (ASHA, 2007).

Limited learning and generalization of speech treatment targets may also reflect procedural learning impairments for children with CAS. One of the most difficult challenges for children with CAS is that, even when provided with a theoretically grounded speech treatment that incorporates principles of motor learning (Maas & Farinella, 2012), some children do not make appreciable gains. In a study that used a single-subject multiple-baselines across-subjects design, Iuzzini and Forrest (2010) tested an intervention that combined

stimulability training and a phonologically modified core vocabulary treatment in four children with CAS (Iuzzini & Forrest, 2010). Results showed that three out of four participants increased their percent consonants correct by 22%–30% following treatment. In contrast, one child increased accuracy by only 9% and showed regression in production of two phonemes. It is not clear why this child—who attended all sessions and had normal cognition—showed a limited response to intervention where the others showed great success. Similarly, another single-subject multiple baseline treatment efficacy study investigated an integral stimulation intervention, Dynamic Temporal and Tactile Cueing treatment. This study showed rapid change in three out of four participants (Strand et al., 2006). Dynamic Temporal and Tactile Cueing currently has one of the strongest evidence bases for treatment of CAS (Maas et al., 2014; Murray & Iuzzini-Seigel, 2017; Murray et al., 2014), yet it is unclear why not all children benefit from it. Procedural learning ability could be an individual factor that impacts response to treatment.

Recent research examined changes to speech motor control during a novel word learning task in 16 children with CAS and typical development (Case & Grigos, 2016). Kinematic analysis was used to track changes to lip and jaw movement during training of two pseudoword exemplars that varied in complexity. Children with CAS showed increased accuracy and consistency; however, they did not show any gains in movement stability, whereas their peers with typical development did. Case and Grigos (2016) suggested that children with CAS may require more practice opportunities to demonstrate changes to their movement stability.

Perhaps the greatest support for the procedural learning deficit hypothesis in children with CAS is provided by studies that examined practice amount or practice distribution. Practice amount refers to the number of practice trials during each session, and practice distribution refers to how these practice trials are divided over time and compares massed practice (many trials in a short period of time) versus distributed practice (same number of practice trials divided over a longer period of time). Edeal and Gildersleeve-Neumann (2011) compared high (100–150 trials/session) and low (30–40 trials/session) practice amount during treatment with integral stimulation treatment. Although both conditions yielded improvements to accuracy, retention and transfer were higher for speech sounds trained in the high practice condition. Likewise, Maas et al. (2019) investigated practice amount and distribution in six children with CAS and found that four of six children showed greater benefit from the high practice and massed practice conditions. Findings suggest that children with CAS require a high number of exposures, over a relatively short period of time to learn and retain speech sound sequences, although again, individual differences prevail. Namasivayam et al. (2015) aimed to investigate practice distribution (once a week vs. twice a week) in a study of 37 children with CAS. Findings showed that massed practice was superior in promoting speech gains, particularly in sentence level intelligibility and Goldman-Fristoe Test of Articulation–Second Edition (GFTA-2) scores;

however, it should be noted that practice amount and distribution were conflated in their design. Both groups completed 10 weeks of treatment such that the massed practice group completed twice as many practice trials as the distributed practice group during the same time period. Taken together, these studies suggest that children with CAS benefit from a higher number of practice trials over a shorter amount of time.

The preceding citations and discussion justify the importance of determining the association among procedural learning, speech, motor, and language production in children with CAS and other communication disorders. This information is critically important to establishing increased understanding of factors that contribute to disorder symptoms, severity, and response to treatment.

### ***Purpose and Research Questions***

The current study is the first to test procedural learning in children with CAS. A customized serial reaction time task, described below, was used to assess procedural learning in these groups. Our well-established diagnostic protocol for differentially diagnosing CAS and other SSDs was used to ensure internal validity and encourage replication. Based on the procedural learning literature in children with DLD and dyslexia and the CAS treatment literature, we posited that impaired procedural learning would be evident in a subset of our participants with CAS and that this would be worse among those with poor grammar.

### **Method**

Thirteen children with CAS and 35 age-matched controls (15 with typical development, 20 with SSD) participated in this prospective case-control study (see Supplemental Material S1 for the STROBE Statement checklist of items to be included in a case-control study). We had aimed to include 20 participants in each group based on our sample size calculation, but the Covid-19 pandemic prematurely halted in-person data collection efforts, required for the current study. Participants were recruited from the Greater Milwaukee area via flyers and referrals from local area clinicians and through social media. Children ranged in age between 43 and 97 months ( $M = 66$  months,  $SD = 12$ ). A subset of 33 participants were included in Iuzzini-Seigel's (2019) investigation of motor performance in children with CAS and other SSDs. Exclusionary criteria included orofacial weakness or craniofacial anomalies, cognitive impairments that prevented participation in experimental procedures and tasks, and hearing impairment. 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. All procedures were approved by the Marquette University Institutional Review Board.

All participants completed a series of communication, cognitive, and motor assessments. 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. Testing includes the Sounds-in-Words subtest of the GFTA-3 (Goldman & Fristoe, 2015), Receptive and Expressive Language components of the Clinical Evaluation of Language Fundamentals–Fifth Edition (CELF-5; Wiig et al., 2013) or Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Wiig et al., 2004), the nonverbal components of the Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003), the Movement Assessment Battery for Children–Second Edition (Movement ABC-2; Henderson et al., 2007), and a customized serial reaction time task designed to assess procedural learning (Nissen & Bullemer, 1987). Two early participants completed the Test of Integrated Language and Literacy Skills (TILLS; Nelson et al., 2016) instead of the CELF. Our lab initially used the TILLS to evaluate language and literacy but then switched to the CELF so that we would have separate, standardized expressive and receptive language index scores. 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, but their language scores were not included in descriptive or statistical analyses. Participants also completed the oral structure and oral function components of the Robbins and Klee (1987) oral mechanism 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. No participants demonstrated evidence of any type of dysarthria. Speech was not assessed as part of this oral function assessment.

### ***Group Assignment***

Children were assigned to groups based on standardized and custom assessments using a well-established protocol in our lab (Iuzzini-Seigel, 2019; Iuzzini-Seigel et al., 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 using the protocol below. This rater was blinded to any previous differential diagnosis participants may have had.

The CAS features that were used to determine group assignment included inconsistency, vowel errors, stress errors, consonant distortions, groping, syllable segregation, intrusive schwa, slow rate, voicing errors, resonance or nasality disturbance, increased difficulty with multisyllabic words, and difficulty in achieving initial articulatory configurations or transitional movement gestures. See Iuzzini-Seigel et al. (2017) or Iuzzini-Seigel and Murray (2017) for operational definitions for each feature and Iuzzini and Forrest (2010) for further explanation of the phonemic inconsistency measure.

**Table 1.** Demographic and speech and language data by participant.

Group	Subgroup	Age (mo.)	Sex	GFTA-3 SS <sup>b</sup>	Average no. of CAS features <sup>c</sup>	Phonemic Inconsistency % <sup>d</sup>	Expressive Language SS <sup>f</sup>	Receptive Language SS <sup>f</sup>
CAS								
001 <sup>a</sup>	CAS-only	58	2	40	5.4	31.7	98	105
002 <sup>a</sup>	CAS-only	73	2	61	5.6 <sup>e</sup>	50.0 <sup>g</sup>	115	105
003	CAS + LI	48	2	40	8	41.5	n/a	75
004	CAS + LI	48	2	40	5.4	34.1	45	90
005	CAS + LI	52	1	40	5.1	33.3	n/a	63
006 <sup>a</sup>	CAS + LI	54	2	40	6.6	35.8	73	67
007 <sup>a</sup>	CAS + LI	56	2	40	5.3	30	63	107
008 <sup>a</sup>	CAS + LI	65	2	40	6.2	34.9	55	77
009 <sup>a</sup>	CAS + LI	68	1	40	5.3	19.5	45	55
010 <sup>a</sup>	CAS + LI	71	1	40	6.3	23.5	63	79
011 <sup>a</sup>	CAS + LI	77	2	40	5.6	29.3	45	45
012 <sup>a</sup>	CAS + LI	77	2	41	5.6	37.4	45	45
013 <sup>a</sup>	CAS + LI	82	1	40	6.9	28.4	45	45
SSD								
014 <sup>a</sup>	SSD-only	43	1	75	3.4	10.6	100	98
015	SSD-only	50	1	73	2.5	7.3	105	113
016 <sup>a</sup>	SSD-only	51	2	59	4	13.0	120	113
017 <sup>a</sup>	SSD-only	54	1	81	2.4	3.3	100	101
018	SSD-only	57	1	72	3	2.4	94	103
019 <sup>a</sup>	SSD-only	57	1	51	4.4	8.1	98	121
020	SSD-only	58	1	62	3.1	6.5	107	119
021	SSD-only	59	2	84	2.5	5.7	92	86
022 <sup>a</sup>	SSD-only	67	2	71	1.9	4.1	108	115
023	SSD-only	68	1	62	2.1	6.5	100	115
024	SSD-only	72	1	70	1.7	1.6	121	115
025	SSD-only	72	1	61	2.5	4.1	96	100
026 <sup>a</sup>	SSD-only	74	2	82	2	1.6	92	92
027 <sup>a</sup>	SSD-only	80	1	54	2.9	0.8		36 <sup>g</sup>
028 <sup>a</sup>	SSD-only	84	2	76	3.8	0.8		31 <sup>g</sup>
029 <sup>a</sup>	SSD + LI	63	2	95 <sup>h</sup>	3.9	0	98	81
030 <sup>a</sup>	SSD + LI	72	2	44	2.6	4.9	52	69
031 <sup>a</sup>	SSD + LI	75	2	75	2.8	3.3	100	81
032 <sup>a</sup>	SSD + LI	92	2	81	2.2	1	85	85
033 <sup>a</sup>	SSD + LI	97	2	40	2.2	7	85	77

(table continues)

Table 1. (Continued).

Group	Subgroup	Age (mo.)	Sex	GFTA-3 SS <sup>b</sup>	Average no. of CAS features <sup>c</sup>	Phonemic Inconsistency % <sup>d</sup>	Expressive Language SS <sup>f</sup>	Receptive Language SS <sup>f</sup>
TD								
034	TD	51	2	101	3	0.8	100	117
035 <sup>a</sup>	TD	51	1	101	3.1	2.4	102	121
036	TD	56	1	105	2.4	1.6	104	98
037	TD	61	2	104	2.9	0	109	107
038	TD	61	2	101	2.1	0	95	98
039 <sup>a</sup>	TD	62	2	105	2.2	0	104	103
040 <sup>a</sup>	TD	65	2	101	2.1	0.8	111	100
041 <sup>a</sup>	TD	72	1	98	2.4	0	116	113
042	TD	72	1	97	1.2	0	111	109
043 <sup>a</sup>	TD	75	2	99	2.3	0	106	111
044 <sup>a</sup>	TD	76	1	114	1.4	0	134	139
045 <sup>a</sup>	TD	76	1	102	1.3	0	132	123
046 <sup>a</sup>	TD	77	2	100	1.2	0.8	120	121
047 <sup>a</sup>	TD	77	1	88	2.2	0	89	109
048 <sup>a</sup>	TD	79	1	93	1.7	0	87	102

Note. mo. = months; CAS = childhood apraxia of speech; LI = language impairment; SSD = speech sound disorder; TD = typically developing.

<sup>a</sup>Participant also participated in Luzzini-Seigel's (2019) study of motor performance in children with typical and disordered communication. <sup>b</sup>Goldman-Fristoe Test of Articulation–Third Edition (GFTA-3; Goldman & Fristoe, 2015). <sup>c</sup>A number of CAS features were elicited in productions of the GFTA-3, customized lists of real 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). <sup>d</sup>Phonemic Inconsistency determined using the Inconsistency Severity Percentage, calculated on whole word responses from the GFTA-3 (Luzzini-Seigel et al., 2017). <sup>e</sup>Participant 002 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%). Lexical inconsistency was evaluated across two productions of the multisyllabic word list and contributed to group assignment. <sup>f</sup>Expressive & Receptive Language standard scores from the Clinical Evaluations of Language Fundamentals Preschool–Second Edition (Wiig et al., 2004) or Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013). N/A was noted for Expressive Language two participants because speech was too limited and severe for accurate and confident scoring. <sup>g</sup>Sum of Identification Core Scores from the Test of Integrated Language & 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 two participants who underwent assessment with this testing instrument. <sup>h</sup>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.



Inconsistency was measured at the phonemic level and was determined using the Inconsistency Severity Percentage (ISP; Iuzzini & Forrest, 2010; Iuzzini-Seigel et al., 2017):

$$\Sigma \left( \frac{\text{(number of different error types-1 for each phoneme)}}{\Sigma(\text{total number of target opportunities}) \times 100.} \right) \quad (1)$$

ISPs of 18% or higher were considered positive for inconsistency (Iuzzini-Seigel, 2019; Iuzzini-Seigel et al., 2017). If a child had an average of five or more CAS features but evidenced an ISP below 18%, their token-to-token inconsistency was assessed using five repetitions of the phrase “buy Bobby a puppy” (Iuzzini-Seigel et al., 2017). If their token-to-token inconsistency of this phrase was higher than 0% (i.e., any of their productions differed from any of their other productions), they were considered positive for the inconsistency feature (Iuzzini-Seigel et al., 2017).

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 et al., 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) or the “Frog Where Are You?” story retell task (Mayer, 1969). The customized word list contained multisyllabic challenging words that ranged from three to four syllables (e.g., synthesis) and build upon words containing stimuli of increasing length, such that triads contained the same root word (e.g., rack, racket, racquetball). These stimuli help to determine if a child is having increased difficulty with multisyllabic words compared with monosyllabic targets. The nonwords contain mono- and multisyllabic exemplars ranging from one to five syllables (Nelson et al., 2016). All words 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 GFTA-3 and multisyllabic word list were each presented twice with a different task in between the two administrations.

Participants were assigned to the CAS group ( $n = 13$ ) if they evidenced a GFTA-3 standard score of  $\leq 85$  and inconsistency and averaged five or more CAS features across the speech tasks (Iuzzini-Seigel, 2019; Iuzzini-Seigel et al., 2017; Iuzzini-Seigel & Murray, 2017). Inclusion in the SSD group ( $n = 20$ ) was based on a GFTA-3 standard score of  $\leq 85$ , no inconsistency,  $< 5/11$  CAS features averaged across speech tasks, and no previous diagnosis or treatment of 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 ( $n = 15$ ) based on a GFTA-3 standard score of  $> 85$ ,  $< 5/11$  CAS features, no inconsistency, 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 TD group, typical language was based on scores of  $> 85$  on the Core Language Score, Receptive Language Score, and Expressive Language Score on the CELF-P2 or CELF-5. The cut score to diagnose language/literacy disorders on the TILLS is 24 for children aged 72–95 months; consequently, typical language abilities were indicated for the two children (both with SSD) 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 two children were used for group assignment but omitted from descriptive and statistical analyses.

### *Other Pertinent Information*

A case history form was completed for each participant by their parent or caregiver. Three participants were reported to have a history of seizures (one child with CAS and normal language reported febrile seizures, one child with CAS and impaired language reported epilepsy, and one child with SSD and normal language reported epilepsy).

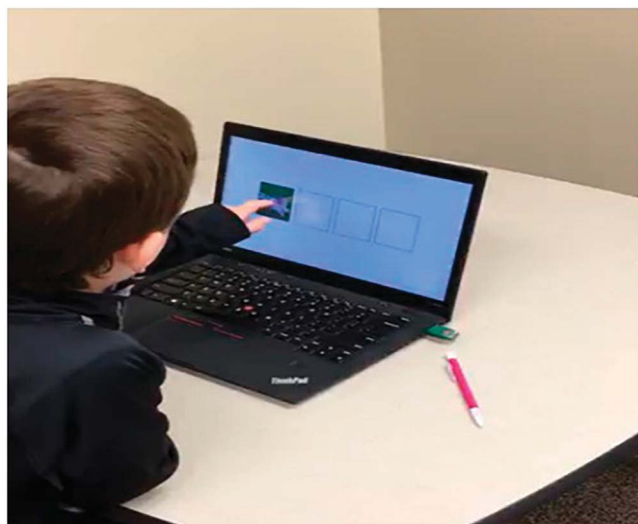
### *Reliability*

A speech pathology student who had completed the rigorous feature rating training in our lab rated CAS features for 15% of participants divided across groups (Iuzzini-Seigel, 2019). Interrater reliability for feature ratings was 92%. Disagreements between raters were resolved through consensus agreement.

### *Procedural Learning Assessment*

We used an adaptation of the classic serial reaction time task (Nissen & Bullemer, 1987) to test procedural learning of a five-step visuospatial sequence. The serial reaction time task is considered the gold standard for procedural learning testing and is easily modified for use by young children. In the current study, participants sat in front of a laptop computer with a touch screen. The computer was fit into a custom wooden frame such that the touch screen was held in a stable position preventing backward movement when the child pressed it to record a response. Four squares were presented across the screen (see Figure 1). The child was instructed to “catch the puppy as fast as you can by touching the screen where the puppy appears.” The left-most square represented Position “1,” the middle two squares represented Positions “2” and “3,” and the right-most square represented Position “4” horizontally. There were two conditions: random and sequenced. For the random condition, the puppy appeared in a random order among the squares on the screen (50 random positional moves/block). For the sequenced condition, the puppy appeared in a five-step sequence (4-2-3-1-2) that repeated 10 times per block (50 sequenced trials/block). The sequenced block was presented 5 times over the course of two sessions (3 times during Session 1 and 2 times during Session 2) for a total of 250 sequenced trials. Sessions occurred within

**Figure 1.** Participant completing the procedural learning serial reaction time task. Responses are recorded via touch screen.



4 days of one another (e.g., Session 1 on a Tuesday and Session 2 on a Friday), similar to a gap one might experience between treatment sessions. Reaction times from the touch screen were recorded by E-Prime.

### Data Processing

Each block of 50 procedural learning trials was divided into 10 bins that each contained five trials. Outliers, defined as any reaction times that exceeded 3 *SDs* above or below the mean reaction time for the block, were removed. This outlier treatment resulted in the retention of 92% of data across conditions. Next, a median reaction time was determined for each bin, and these medians were averaged across bins. Group performance on reaction time of sequenced trials was evaluated across blocks and sessions and relative to random trials. If a group evidenced procedural learning, reaction time would be expected to decrease across each sequenced block, then increase on the penultimate block, which contained random trials, and then decrease again on the final sequenced block.

### Motor Testing

All participants underwent fine and gross motor testing using the Movement ABC-2 (Henderson et al., 2007), which yields component scores for manual dexterity, balance, and aiming and catching. The Movement ABC-2 is sensitive at detecting even mild motor impairments (Van

Waelvelde et al., 2007) in children with a range of cognitive abilities (Henderson & Sugden, 1992; Spanò et al., 1999; Sugden & Wann, 1987). Our previous research (Iuzzini-Seigel, 2019) reported the results of this assessment on the motor abilities on 33 of the children included in the current study and showed that the CAS group performed below the normal limit on all components of the Movement ABC-2 assessment, whereas the TD children and those with SSD performed within the normal range, on average. In addition, children with CAS performed significantly poorer than those with typical development and SSD on the Aiming and Catching and Balance subtests. See Iuzzini-Seigel (2019) for an in-depth description of motor tests and resultant findings.

### Statistical Treatment

Statistical assumptions were assessed for each variable, and nonparametric tests were used where necessary. Missing data cases were excluded pairwise from analyses. In total, 4 data points were missing from the Manual Dexterity subtest, 1 from Aiming and Catching, and 1 from Balance. Two children had missing data on the CELF as they were administered the TILLS. An additional two children had missing data on the Expressive Portion of the CELF, due to speech severity. Two children had missing data on the Reynolds Intellectual Assessment Scales. Two children had missing data on the Oral Structure assessment; and three children, on the Oral Function assessment. Three children had missing data on the Session 2 procedural learning assessment. Analyses of variance (ANOVAs) or Kruskal-Wallis tests were used to detect group differences for age, articulation, expressive language, receptive language, oral mechanism structure and oral function scores, nonverbal IQ scores, and Movement ABC-2 component scores for Aiming and Catching, Manual Dexterity, and Balance. Bonferroni-corrected post hoc *t* tests or Mann-Whitney *U* tests were used to identify pairwise differences and control for familywise error rate. Within- and between-group differences for procedural learning of random and sequenced reaction times were identified using mixed ANOVAs. Interactions between Group  $\times$  Condition and Group  $\times$  Time were also examined. After data were analyzed by group, data were reanalyzed based on grammatical ability from performance on the Sentence Structure or Sentence Comprehension, Word Structure, and Recalling Sentences subtests of the CELF assessments. This was based on a protocol established by Hedenius et al. (2011). Sentence Structure (for CELF-P2) and Sentence Comprehension (for CELF-5) evaluate the ability to interpret spoken sentences of increasing length and complexity. Word Structure evaluates knowledge of grammatical rules in a sentence completion task. Recalling Sentences evaluates the child's ability to remember and reproduce sentences of increasing length and complexity while maintaining correct content, morphology, and syntax. If the child scored below 7 on two or more of these subtests, they were assigned to the disordered grammar group.

## Results

### Demographics

ANOVAs or Kruskal–Wallis tests were used to detect group differences in demographic, speech, language, oral mechanism, and cognitive variables. Kruskal–Wallis tests and Mann–Whitney *U* tests were used when assumptions of homogeneity of variance or normality were not met. See Table 2 for a summary of data by group, including notation of statistically significant differences between groups.

No significant group difference was found for age,  $F(2, 45) = 0.39, p = .678, \eta_p^2 = .004$ . An ANOVA showed a main effect of group for nonverbal IQ,  $F(2, 43) = 4.57, p = .016, \eta_p^2 = .167$ , where the CAS group scored significantly lower than the TD group ( $p = .013$ ). *t* Kruskal–Wallis tests revealed a group difference for oral function scores on the oral mechanism assessment,  $H(2) = 17.70, p = .001$ , where the CAS group performed more poorly than the TD ( $p < .001$ ) and SSD ( $p = .025$ ) groups; no other group differences for oral function were detected. No group effect for oral structure was found ( $p = .213$ ). A main group effect

**Table 2.** Demographic and speech/language data by speech group.

Variable	Group		
	CAS ( <i>n</i> = 13)	SSD ( <i>n</i> = 20)	TD ( <i>n</i> = 15)
Demographic measures			
Age in months	64 (12)	67 (14)	67 (10)
Sex	4F, 9M	10F, 10M	8F, 7M
Nonverbal IQ SS	96 (28) <sub>a</sub>	110 (15)	119 (16) <sub>a</sub>
Speech measures			
Articulation SS	42 (6) <sub>a,b</sub>	68 (14) <sub>a,c</sub>	101 (6) <sub>b,c</sub>
CAS Features	6 (0.8) <sub>a,b</sub>	3 (0.8) <sub>a</sub>	2 (0.6) <sub>b</sub>
Inconsistency Severity %	32 (6) <sub>a,b</sub>	5 (3) <sub>a,c</sub>	0.4 (0.7) <sub>b,c</sub>
Language measures			
Expressive Language SS	63 (24) <sub>a,b</sub>	97 (15) <sub>a</sub>	108 (14) <sub>b</sub>
Receptive Language SS	74 (23) <sub>a,b</sub>	99 (16) <sub>a</sub>	111 (11) <sub>b</sub>
Oral mechanism measures			
Oral structure score	23 (1)	23 (2)	24 (1)
Oral function score	26 (5) <sub>a,b</sub>	31 (1) <sub>a</sub>	32 (1) <sub>b</sub>

*Note.* Group averages listed with standard deviations in parentheses. Groups sharing the same subscript letter were statistically different for the specified variable. CAS Features: Iuzzini-Seigel et al., 2017. Inconsistency Severity %: Iuzzini & Forrest, 2010. Expressive Language and Receptive Language SS are from Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Wiig et al., 2004) or Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013) for participants older than 6 years of age. Oral mechanism “oral structure score” is from Robbins and Klee (1987); The highest possible score is 24, and scores are expected to be 20–24 for this age range. Oral mechanism “oral function score” is from Robbins and Klee (1987); The highest possible score is 32; no age norms are available for this measure. CAS = childhood apraxia of speech; SSD = speech sound disorder; TD = typically developing; F = female; M = male; SS = standard score; nonverbal IQ = Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003); Articulation SS = Goldman-Fristoe Test of Articulation–Third Edition (Goldman & Fristoe, 2015).

was also found for expressive,  $H(2) = 17.26, p < .001$ , and receptive,  $H(2) = 17.14, p < .001$ , language. The CAS group had poorer expressive language ( $M = 63, SD = 24$ ) than the TD ( $p < .001; M = 108, SD = 24$ ) and SSD ( $p = .019; M = 98, SD = 15$ ) groups. Post hoc Bonferroni-adjusted pairwise tests revealed that children with CAS also evidenced significantly lower receptive language than children with SSD ( $p = .021$ ) and typical development ( $p < .001$ ). On average, children with CAS had an average receptive language standard score of 74 ( $SD = 23$ ), while the SSD group averaged 100 ( $SD = 16$ ), and the TD group averaged 111 ( $SD = 11$ ). Kruskal–Wallis tests revealed a significant group effect for GFTA-3 standard scores,  $H(2) = 39.60, p < .001$ , and the ISP,  $H(2) = 37.07, p < .001$ . Note that significant group differences in speech assessments are expected as these were the basis of group assignment. Children with CAS evidenced significantly lower standard scores on the GFTA-3 compared to children with SSD ( $p = .007$ ) and typical development ( $p < .001$ ); children with SSDs scored lower than children with typical development as well ( $p = .001$ ). On average, children with CAS had standard scores of 42 ( $SD = 6$ ), those with SSD had average scores of 68 ( $SD = 15$ ), and those with typical development averaged 101 ( $SD = 6$ ). Children with CAS evidenced significantly higher ISP scores relative to children with SSD ( $p = .004$ ) and typical development ( $p < .001$ ); children with SSD evidenced higher ISPs relative to children with typical development as well ( $p = .002$ ). On average, children with CAS evidenced an ISP of 32 ( $SD = 6$ ), while the SSD group had an ISP of 5 ( $SD = 3$ ), and the TD group had an ISP of  $< 1$  ( $SD = 1$ ). Kruskal–Wallis tests revealed a main effect of group for an average number of CAS features,  $H(2) = 31.21, p < .001$ , wherein the CAS group evidenced more features ( $M = 6, SD = 0.8$ ) than the TD ( $M = 2, SD = .6$ ) and SSD ( $M = 3, SD = 0.8; p < .001$ ) groups no significant difference was found between the SSD and TD groups.

### Motor Performance

The assumption of normality was not met for the Movement ABC-2 subtests. As such, Kruskal–Wallis tests were used to determine group differences on the Manual Dexterity,  $H(2) = 7.31, p = .026$ ; Aiming and Catching,  $H(2) = 13.92, p = .001$ ; and Balance,  $H(2) = 15.73, p < .001$ , test components. Bonferroni-adjusted post hoc tests were used to detect pairwise differences. On Manual Dexterity, children with CAS performed significantly more poorly than the SSD group ( $p = .043$ ) but not the TD group ( $p = .058$ ) once the Bonferroni correction was made. For the movement component scores, the manual considers a 5 or below to be the “red zone,” reflecting significant movement difficulty that likely needs therapeutic intervention. On average, for Manual Dexterity, children with CAS scored 4 ( $SD = 4$ ) compared to those with SSD who scored 7 ( $SD = 6$ ) and typical development who scored 8 ( $SD = 4$ ). On Aiming and Catching, the CAS group (average = 4,  $SD = 3$ ) performed significantly more poorly than the SSD ( $p = .024$ ) and TD ( $p = .001$ ) groups who scored 8 ( $SD = 3$ ) and 10 ( $SD = 3$ ), respectively. Finally, on the Balance component,

children with CAS performed more poorly than the SSD ( $p = .001$ ) and TD ( $p = .003$ ) groups. The CAS group scored 4 ( $SD = 3$ ), while the SSD group scored 9 ( $SD = 3$ ), and the TD group scored 8 ( $SD = 2$ ). See Figure 2 for motor performance data presented by group.

### Procedural Learning

The procedural learning assessment was designed such that a child had to choose the correct square in order to progress to the subsequent trial; this was done to prevent the child from arbitrarily pressing all over the screen to complete the assessment as quickly as possible. If the child touched the area outside of the target square, no change occurred on the screen—the puppy stayed in the same position. Consequently, during analysis, 100% of responses were considered correct, but reaction times could be considerably longer if a child initially touched outside the target square. Assumptions were met to conduct parametric testing on procedural learning data. Reaction times of sequenced and random trials were analyzed.

#### Reaction Time of Sequenced and Random Trials During Session 1

Reaction times for the bin medians for the three sequenced blocks were averaged together as were the bin medians for the two random blocks. These means were compared within and between groups in a mixed ANOVA. Results showed a main effect of condition (random vs. sequenced),  $F(1, 45) = 14.01, p = .001, \eta_p^2 = .237$ , and group,  $F(2, 45) = 17.30, p < .001, \eta_p^2 = .435$ , but no significant Condition  $\times$  Group interaction. Post hoc tests showed that the CAS group evidenced significantly slower reaction time on random and sequenced trials (916 and 911 ms,

respectively) compared with the TD (random = 708 ms, sequenced = 675 ms) and SSD (random = 688 ms, sequenced = 651 ms) groups ( $p < .001$ ). Descriptive data from Session 1 showed that, on average, the CAS group was 5 ms faster on sequenced trials relative to random trials. In contrast, the TD group was 33 ms faster on sequenced trials, and the SSD group was 37 ms faster on this condition.

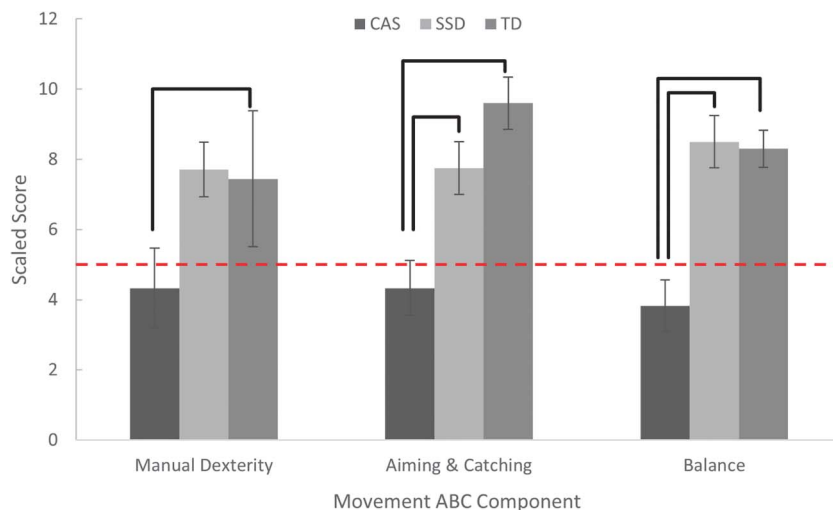
#### Reaction Time of Sequenced and Random Trials During Session 2

Data from Session 2 were analyzed using a mixed ANOVA that compared average reaction time on sequenced trials with random trials, within and between groups. Results showed significant effects for condition,  $F(1, 42) = 13.32, p = .001, \eta_p^2 = .241$ , where random trials were longer than sequenced trials, and group,  $F(2, 42) = 9.08, p = .001, \eta_p^2 = .302$ , where the CAS group was slower on both random and sequenced trials (random = 835 ms, sequenced = 798 ms) compared with the SSD (random = 621 ms, sequenced = 600 ms) and TD (random = 656 ms, sequenced = 607 ms) groups. During Session 2, the CAS group was 37 ms faster on sequenced relative to random trials while the TD and SSD groups were 49 and 21 ms, respectively, faster on sequenced trials relative to random trials. Again, no Group  $\times$  Condition interaction was detected.

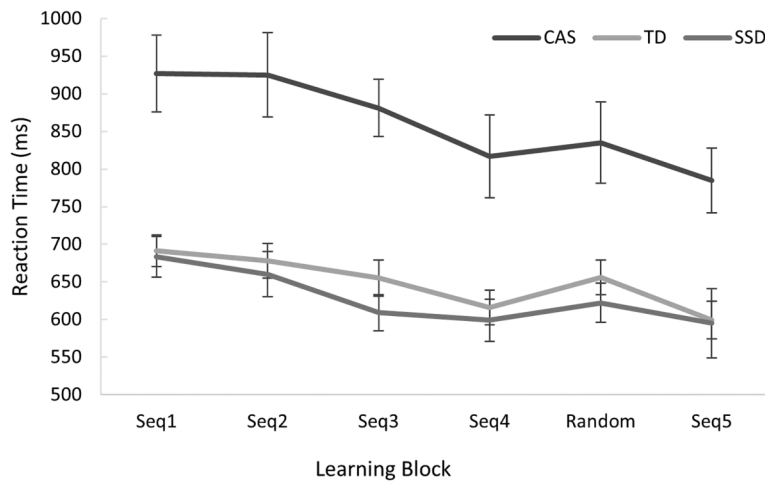
#### Within- and Between-Group Differences Across Five Sequenced Blocks

Next, a mixed ANOVA was used to determine within- and between-group differences on reaction time across the five sequenced blocks. See Figure 3 for procedural learning data across blocks for speech groups. Results showed a main effect of time,  $F(4, 168) = 22.14, p < .001, \eta_p^2 = .345$ , in

**Figure 2.** Movement ABC-2 component scores by group. The red line marks the cutoff for the “red zone” wherein a score of 5 or below indicates 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 = typically developing.



**Figure 3.** Procedural learning data across blocks for speech groups. A rebound effect (i.e., uptick in reaction time) during the random block indicates that procedural learning occurred. CAS = childhood apraxia of speech; SSD = speech sound disorder; TD = typically developing.



which reaction time decreased on sequenced trials over time, and group,  $F(2, 42) = 14.64, p < .001, \eta_p^2 = .411$ , where the CAS group was slower on all blocks compared to other groups; no Group  $\times$  Time interaction was detected. Descriptive data showed that the median reaction time for the CAS group increased at sequence Block 2, whereas the other groups showed decreased reaction time during each subsequent sequenced block.

Next, as an indication of whether procedural learning occurred by the end of the serial reaction time task, we determined how many children in each group demonstrated an uptick in reaction time on the random block that occurred in between the two final sequenced blocks during Session 2. Using this method and including only children with complete procedural learning data, 7/11 (63%) children with CAS demonstrated procedural learning as did 13/15 (87%) TD children and 13/19 (68%) children with SSD.

### Demographics for Groups Assigned Based on Grammatical Ability

Children were reassigned to groups based on grammatical ability. Grammatical ability was determined based on performance on the Word Structure, Recalling Sentences, and Sentence Structure/Sentence Comprehension subtests on the CELF-P2 and CELF-5, using a similar methodology to that used by Hedenius et al. (2011). If a child scored below 7 on two or more of these subtests, they were assigned to the impaired grammar group. This resulted in 12 children being assigned to the impaired grammar group and 36 children being assigned to the typical grammar group. Of the 12 children with poor grammar, 11 had CAS and one had SSD. Mann-Whitney  $U$  tests were used to detect group differences in demographic, communication, oral function, and motor variables.

Results revealed significant differences between groups ( $p < .001$ ) for all speech, language, oral function, and motor

variables, where the impaired grammar group performed more poorly relative to the typical grammar group. No significant group differences were detected for age ( $p = .489$ ), nonverbal IQ ( $p = .072$ ), or oral structure ( $p = .075$ ). See Table 3 for summary data for groups assigned on grammatical ability.

### Procedural Learning in Children Reclassified on Basis of Grammatical Ability

A mixed ANOVA evaluated within- and between-group differences of reaction time for the random and sequenced blocks from Session 1. Findings revealed a main effect of condition,  $F(1, 46) = 6.96, p = .011, \eta_p^2 = .131$ , where sequenced trials were faster than random trials, and a main effect for group,  $F(1, 46) = 19.52, p < .001$ , where children with TD grammar evidenced faster reaction time on random and sequenced trials relative to children with impaired grammar. There was also a marginal Group  $\times$  Condition interaction,  $F(1, 46) = 3.91, p = .054, \eta_p^2 = .078$ , where the impaired grammar group showed no effect of condition (sequenced or random) while the typical grammar group did.

Next, data from Session 2 were analyzed using a mixed ANOVA that compared average reaction time of sequenced and random trials. Results showed significant effects for condition,  $F(1, 43) = 6.85, p = .012, \eta_p^2 = .137$ , where random trials were longer than sequenced trials, and group,  $F(1, 43) = 13.75, p < .001, \eta_p^2 = .242$ , where the impaired grammar group was slower on random and sequenced trials compared with the typical grammar group. No Group  $\times$  Condition interaction was detected.

Finally, a mixed ANOVA was used to determine within- and between-group differences of reaction time for the five sequenced trials. See Figure 4 for procedural learning data across blocks for grammatical groups. Results showed a main effect of time,  $F(4, 172) = 21.38, p < .001, \eta_p^2 = .322$ , where reaction time decreased over time with

**Table 3.** Demographic and speech/language data for groups assigned on grammatical ability.

Variable	Group	
	Impaired grammar ( <i>n</i> = 12)	Typical grammar ( <i>n</i> = 36)
Demographics		
Age in months	64 (12)	67 (12)
Sex	4F, 8M	18F, 18M
Nonverbal IQ SS	96 (30)	114 (16)
Speech measures		
Articulation SS	40 (1) <sub>a</sub>	82 (20) <sub>a</sub>
CAS Features	6 (1) <sub>a</sub>	3 (1) <sub>a</sub>
Inconsistency Severity %	29 (10) <sub>a</sub>	4 (6) <sub>a</sub>
Language measures		
Expressive Language SS	53 (10) <sub>a</sub>	104 (12) <sub>a</sub>
Receptive Language SS	68 (19) <sub>a</sub>	106 (14) <sub>a</sub>
Oral mechanism measures		
Oral structure score	23 (1)	23 (1)
Oral function score	27 (5) <sub>a</sub>	31 (2) <sub>a</sub>
Motor measures		
Manual Dexterity	3 (2) <sub>a</sub>	8 (3) <sub>a</sub>
Aiming and Catching	4 (3) <sub>a</sub>	9 (3) <sub>a</sub>
Balance	3 (2) <sub>a</sub>	8 (3) <sub>a</sub>

*Note.* Group averages are listed with standard deviations in parentheses. Groups sharing the same subscript letter were statistically different for the specified variable. CAS Features: luzzini-Seigel et al., 2017. Inconsistency Severity %: luzzini & Forrest, 2010. Expressive Language and Receptive Language SS are from Clinical Evaluation of Language Fundamentals–Second Edition (Wiig et al., 2004) or Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013) for participants older than 6 years of age. Oral mechanism “oral structure score” is from Robbins and Klee (1987): The highest possible score is 24, and scores are expected to be 20–24 for this age range. Oral mechanism “oral function score” is from Robbins and Klee (1987): The highest possible score is 32; no age norms are available for this measure. Manual Dexterity, Aiming and Catching, and Balance are component scaled scores from the Movement Assessment Battery for Children–Second Edition (Henderson et al., 2007). F = female; M = male; SS = standard score; CAS = childhood apraxia of speech; nonverbal IQ = Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003); Articulation SS = Goldman-Fristoe Test of Articulation–Third Edition (Goldman & Fristoe, 2015).

practice, and group,  $F(1, 43) = 25.94, p < .001, \eta_p^2 = .376$ , where the impaired grammar group was slower than the typical grammar group. Finally, in contrast to the results when we analyzed data by speech diagnosis, we also observed a Group  $\times$  Time interaction,  $F(4, 172) = 2.68, p = .033, \eta_p^2 = .059$ . For the impaired grammar group, data showed that reaction time, on average, increased at sequenced Block 2, where the typical grammar group showed decreased reaction time during each sequenced block.

### **Post Hoc Profile of Children Grouped by Procedural Learning Pattern**

A visual scan of the data revealed that a subset of participants in each group increased their reaction time during sequenced Block 2, rather than decreasing it on each successive block. Because of this, we decided to reassign children

to groups based on whether or not they displayed this pattern, resulting in 18 children being assigned to the increased reaction time during Block 2 (Inc\_RT) group and 30 children being assigned to the decreased reaction time during Block 2 (Dec\_RT) group. Nonparametric Mann–Whitney *U* tests were run to determine group differences in demographic and experimental variables of interest. No group differences were detected for age ( $p = .924$ ). A group difference ( $p = .036$ ) was found for nonverbal IQ where the Inc\_RT group scored 102 ( $SD = 21$ ) and the Dec\_RT group scored 114 ( $SD = 20$ ). A significant group difference was detected for the Manual Dexterity component ( $p = .002$ ), where the Inc\_RT group averaged a scaled score of 4 ( $SD = 2$ ) in contrast to the Dec\_RT group that averaged an 8 ( $SD = 4$ ). No other group differences were detected on speech, language, oral mechanism, or motor tasks when groups were differentiated based on this procedural learning pattern. See Table 4 for summary data for groups assigned based on procedural learning pattern (i.e., did participants increase or decrease their reaction time during sequenced Block 2 relative to their reaction time during Block 1).

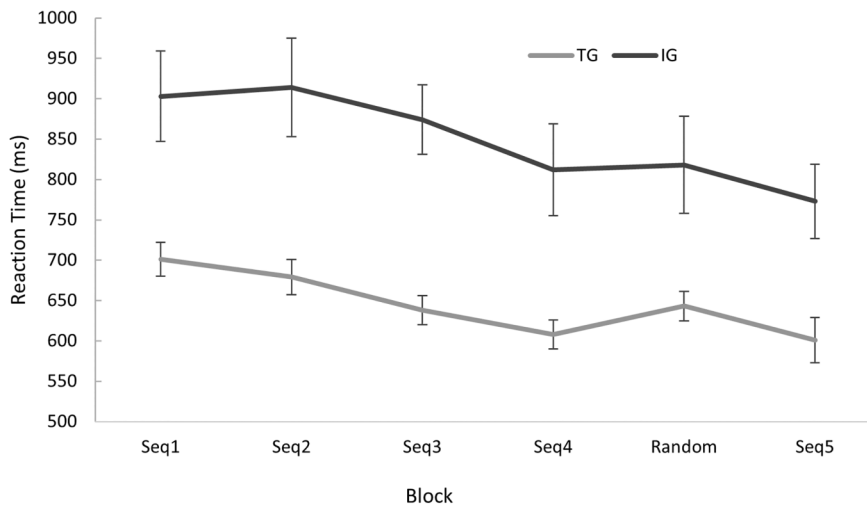
We then assessed group differences on reaction time during the initial and final sequenced blocks. No group difference was detected during the initial sequenced block ( $p = .932$ ; Inc\_RT Group = 778 ms, Dec\_RT Group = 734 ms), but there was a significant group difference detected during the final sequenced block ( $p = .031$ ) for which the Inc\_RT group averaged 735 ms and the Dec\_RT group averaged 587 ms. See Figure 5 for procedural learning data across blocks for groups assigned on procedural learning pattern.

### **Composition of the Inc\_RT and Dec\_RT Groups**

The Inc\_RT group, which demonstrated an uptick in reaction time during sequenced Block 2 relative to Block 1, included six children with CAS, five with typical development, and seven with SSD such that approximately half of the children with CAS were in this group compared with one third of children with typical development and approximately one third with SSD. Descriptive data were examined to identify any trends. See Table 5 for descriptive data for groups assigned on procedural learning pattern and speech diagnosis. For participants with CAS, the Inc\_RT subgroup tended to evidence poorer Receptive and Expressive Language standard scores and scaled scores on the grammatical subtests relative to the Dec\_RT group. For participants with SSD, the Inc\_RT group evidenced poorer performance on Receptive and Expressive Language and on the Manual Dexterity and Aiming and Catching components relative to the Dec\_RT group, yet they also scored higher on the GFTA-3. TD participants with the Inc\_RT pattern tended to have lower scores (even if within the normal range) on Expressive Language, Manual Dexterity, and Aiming and Catching relative to peers with the Dec\_RT pattern.

Reaction time during the initial and final sequenced blocks was explored for each of these groups. Children in the CAS Inc\_RT and Dec\_RT subgroups did not show

**Figure 4.** Procedural learning data across blocks for grammar groups. A rebound effect (i.e., uptick in reaction time) during the random block indicates that procedural learning occurred. TG = typically developing grammar; IG = impaired grammar.



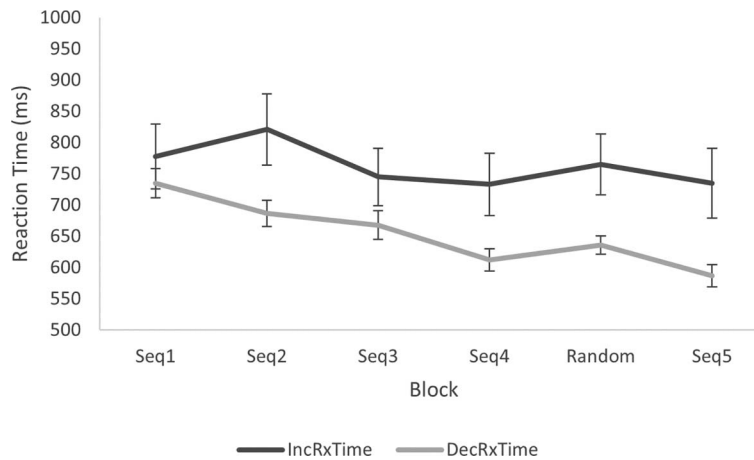
**Table 4.** Demographic and speech/language data for groups assigned on procedural learning pattern.

Variable	Group	
	Increased reaction time (n = 18)	Decreased reaction time (n = 30)
Demographics		
Age in months	66 (12)	67 (13)
Sex	8F, 10M	14F, 16M
Nonverbal IQ SS	102 (21) <sup>a</sup>	114 (19) <sup>a</sup>
Speech measures		
Articulation SS	69 (26)	73 (25)
CAS Features	4 (2)	3 (2)
Inconsistency Severity %	12 (14)	9 (13)
Language measures		
Expressive Language SS	84 (26)	97 (23)
Receptive Language SS	88 (24)	101 (21)
Oral mechanism measures		
Oral structure score	23 (1)	23 (1)
Oral function score	29 (4)	30 (3)
Motor measures		
Manual Dexterity	4 (2) <sup>a</sup>	8 (4) <sup>a</sup>
Aiming and Catching	6 (4)	8 (3)
Balance	7 (3)	8 (3)

*Note.* Participants who demonstrated slower reaction time during sequenced Block 2 relative to their reaction time during Block 1 were assigned to the increased reaction time group, and those who demonstrated faster reaction time during sequenced Block 2 relative to Block 1 were assigned to the decreased reaction time group. Group averages are listed with standard deviations in parentheses. Groups sharing the same subscript letter were statistically different for the specified variable. CAS Features: Iuzzini-Seigel et al., 2017. Inconsistency Severity %: Iuzzini & Forrest, 2010. Expressive Language and Receptive Language SS are from Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Wiig et al., 2004) or Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013) for participants older than 6 years of age. Oral mechanism “oral structure score” is from Robbins and Klee (1987); The highest possible score is 24, and scores are expected to be 20–24 for this age range. Oral mechanism “oral function score” is from Robbins and Klee (1987); The highest possible score is 32; no age norms are available for this measure. Manual Dexterity, Aiming and Catching, and Balance are component scaled scores from the Movement Assessment Battery for Children–Second Edition (Henderson et al., 2007). F = female; M = male; CAS = childhood apraxia of speech; SS = standard score; nonverbal IQ = Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003); Articulation SS = Goldman-Fristoe Test of Articulation–Third Edition (Goldman & Fristoe, 2015).

<sup>a</sup>Statistically significant difference between groups.

**Figure 5.** Procedural learning data across blocks for groups assigned based on procedural learning pattern (i.e., whether reaction time increased or decreased during sequenced Block 2 relative to sequenced Block 1). A rebound effect (i.e., uptick in reaction time) during the random block indicates that procedural learning occurred.



any apparent differences in pattern learning outcomes, although the Inc\_RT group tended to be slower overall; both groups decreased their reaction time by roughly the same amount between sequenced Blocks 1 and 5 (~140 ms). The

SSD and TD groups showed more distinct procedural learning outcomes based on their subgroupings. The Inc\_RT SSD group showed longer reaction time during sequence Block 5 (680 ms) relative to their performance during Block 1

**Table 5.** Descriptive data for groups assigned based on speech diagnosis and procedural learning pattern.

Variable	Group					
	CAS		SSD		TD	
	Inc RT (n = 5)	Dec RT (n = 7)	Inc RT (n = 7)	Dec RT (n = 13)	Inc RT (n = 5)	Dec RT (n = 10)
Age in months	64 (14)	64 (10)	65 (11)	68 (16)	68 (12)	67 (9)
Speech measures						
Articulation	40 (0)	43 (8)	73 (17)	66 (12)	98 (5)	102 (6)
CAS Features	6 (1)	6 (1)	3 (1)	3 (1)	2 (1)	2 (1)
ISP	31 (5)	32 (8)	4 (4)	5 (4)	1 (1)	0 (1)
Language measures						
Receptive Language	65 (18)	81 (25)	92 (15)	104 (16)	112 (10)	111 (13)
Expressive Language	54 (13)	70 (30)	93 (21)	100 (10)	102 (12)	111 (14)
Motor measures						
Manual Dexterity	4 (3)	5 (5)	5 (2)	8 (3)	4 (2)	10 (3)
Aiming and Catching	5 (3)	4 (3)	6 (5)	9 (2)	8 (3)	11 (3)
Balance	3 (2)	4 (3)	8 (3)	9 (3)	8 (2)	9 (2)
Block						
Seq1 RT	991 (234)	872 (120)	657 (134)	697 (120)	694 (109)	689 (68)
Seq2 RT	1,052 (224)	815 (104)	693 (179)	642 (109)	721 (121)	657 (61)
Seq3 RT	945 (147)	826 (109)	628 (144)	599 (87)	671 (102)	647 (90)
Seq4 RT	894 (219)	725 (61)	641 (177)	579 (91)	650 (117)	598 (70)
Random RT	926 (206)	726 (27)	660 (163)	604 (83)	700 (129)	634 (61)
Seq5 RT	848 (165)	710 (550)	680 (329)	557 (95)	665 (131)	566 (60)

*Note.* Participants who demonstrated slower reaction time during sequenced Block 2 relative to their reaction time during Block 1 were assigned to the “Inc RT” group, and those who demonstrated faster reaction time during sequenced Block 2 relative to Block 1 were assigned to the “Dec RT” group. Group averages with standard deviations are listed in parentheses. All speech and language measures presented as standard scores). Articulation = Average standard score on the GFTA-3 (Goldman & Fristoe, 2015). CAS Features: Iuzzini-Seigel et al., 2017. Expressive Language and Receptive Language are from Clinical Evaluation of Language Fundamentals Preschool–Second Edition (Wiig et al., 2004) or Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013) for participants older than 6 years of age. Manual Dexterity, Aiming and Catching, and Balance are component scaled scores from the Movement Assessment Battery for Children–Second Edition (Henderson et al., 2007). CAS = childhood apraxia of speech; SSD = speech sound disorder; TD = typically developing; Articulation = Goldman-Fristoe Test of Articulation–Third Edition (Goldman & Fristoe, 2015); ISP = Inconsistency Severity Percentage (Iuzzini & Forrest, 2010); SeqX RT = sequenced block # reaction time in milliseconds; Random RT = random block reaction time in milliseconds.



(657 ms); in contrast, their peers with the Dec\_RT pattern showed substantially decreased reaction time at Block 5 (557 ms) relative to Block 1 (697 ms). Children in the TD group with the Inc\_RT pattern showed a small decrease in their reaction time at Block 5 relative to Block 1 (~30 ms), whereas the Dec\_RT pattern group showed a more substantial change during this time span (~125 ms).

## Discussion

The extant literature on children with CAS reports a high rate ( $\geq 50\%$ ) of co-occurring motor and cognitive-linguistic impairments in this population (Iuzzini-Seigel, 2019; Lewis et al., 2004; Tükel et al., 2015). It is unknown, however, what links these co-occurring language and motor deficits with the speech features that are central to the CAS diagnosis. The current study tested the procedural learning deficit hypothesis that has been used to explain co-occurring motor, linguistic, literacy, and attentional deficits in other disordered populations such as children with DLD, dyslexia, and attention-deficit/hyperactivity disorder (Nicolson & Fawcett, 2007); to our knowledge, this study represents the first to test this hypothesis in children with CAS. We posited that children with CAS, and those with poor grammar, would evidence poorer procedural learning ability on a serial reaction time task compared to children with SSD and typical development. Findings from the current study partially support this hypothesis.

Over the course of the procedural learning protocol, all groups (but not all individuals) demonstrated decreased reaction time on the sequenced blocks and a rebound effect on the random trials that were presented as the penultimate block, an indication that all groups experienced some amount of procedural learning during this time span. The CAS group, however, exhibited an interesting pattern that was distinct from the average performance demonstrated by the other groups. On average, the CAS group displayed slower reaction time during the second block of sequenced trials relative to their reaction time during the first block; this contrasts with the other groups who showed faster reaction time on each successive block of sequenced trials. After this brief uptick in reaction time, the CAS group decreased reaction time on sequenced trials during the third sequenced block in Session 1 and over the two blocks in Session 2. These results importantly demonstrate that children with CAS, on average, do demonstrate procedural learning but that they require a greater number of exposures to a sequence to demonstrate this learning compared to peers with SSD or typical development (i.e., learning was demonstrated by Blocks 3–4 rather than by Block 2); although children with CAS demonstrated learning, their learning process appeared to differ from control groups of children with SSD and typical development. This preliminary evidence may help to explain why so many children with CAS require highly intense and frequent treatment sessions with hundreds or even thousands of practice trials in order to learn and generalize treatment targets, whereas children with other disorders or typical development are

able to benefit from less intense and less frequent training (e.g., Case & Grigos, 2016; Edeal & Gildersleeve-Neumann, 2011; Maas et al., 2014; Thomas et al., 2014). Previous research that compared moderate- versus high-frequency production practice (i.e., 30 vs. 150 trials per session) showed greater learning and generalization from high-frequency practice in children with CAS (Edeal & Gildersleeve-Neumann, 2011). Other research provides support for more frequent treatment sessions as well (Namasivayam et al., 2015; Thomas et al., 2014). The sequencing required to produce a combination of speech sounds is substantially more complicated than what is required to learn and respond to the visuospatial sequence presented in our five-step serial reaction time task; given that it took more than 20–30 repetitions of the simple sequence to demonstrate learning in the current serial reaction time task, we can extrapolate how many trials might be needed for a child with CAS to demonstrate learning of speech sequences. Future research should aim to more fully ascertain procedural learning ability in larger groups of children with CAS who vary on co-occurring conditions to better understand the extent to which procedural learning deficits affect this population. We should then determine the most efficient treatment schedule and most optimal number of practice trials to facilitate substantive speech gains in children with CAS who demonstrate less efficient procedural learning abilities.

### *Procedural Learning in Groups Assigned Based on Grammatical Ability*

After conducting initial comparisons for groups assigned based on speech diagnosis, we reassigned participants to groups based on grammatical ability using a method adapted from Hedenius et al. (2011). Interestingly, 11 out of 12 children in the impaired grammar group had CAS. It should be noted that there were five additional children with SSD who demonstrated CELF scores in the disordered range but who were not assigned to the impaired grammar group based on their specific profile of language deficits (i.e., mild receptive language deficits with Word Structure scores in the normal range). When participants were assigned to groups based on grammatical ability, the increased reaction time (uptick pattern) that was evidenced by the CAS group during the second sequenced block did result in a significant Group  $\times$  Time interaction. Our findings differ from work by Hedenius et al. who examined procedural learning in children with DLD who were assigned to groups based on grammatical ability. Hedenius et al. found that, while both groups demonstrated initial sequence learning, the impaired grammar group did not show consolidation, which was based on a lack of improvement in reaction time between the last block of trials in Session 1 and the block of trials in Session 2. The current findings did show consolidation across sessions for both grammar groups. The impaired grammar group, however, showed a very small rebound effect on the random trials during the penultimate block, which brings into question the extent to which our participants with impaired grammar actually did benefit from procedural

learning. Children with CP are also known to get faster on this type of task but fail to show a rebound effect in response to introduction of a novel sequence during the penultimate block (Gofer-Levi et al., 2013). Gofer-Levi et al. suggest that this pattern reflects explicit learning of instructions and response to feedback but not implicit sequence learning. In the current study, no feedback was given and there was a small rebound effect during the random trials and then an even greater decrease during the final block of sequenced trials, suggesting that procedural learning did occur in the grammar impaired group, even if this was not a robust effect. The current study should be replicated in larger numbers of children with grammatical deficits, including those with DLD and other SSDs to further disambiguate these findings and determine the extent to which procedural learning occurs in children with grammatical impairments and various comorbid diagnoses.

It is notable that the two grammar groups significantly differed on motor abilities as well, and the impaired grammar group scored in the “red zone” indicating poor performance on all components (Manual Dexterity, Aiming and Catching, and Balance), compared to the typical grammar group who scored in the normal range on all components. These findings provide support for a subgroup (largely composed of children with CAS) who has weaker procedural learning abilities and multisystem speech, language, and motor deficits.

### ***Nonmonotonic Procedural Learning***

Approximately half of the participants with CAS and one third of those with typical development and SSD evidenced an uptick in their reaction time during the second block of sequenced trials, an unexpected finding. To learn more about the children who showed this pattern, we examined descriptive data for each of these groups. The most robust takeaway was that, although the two groups started out with comparable reaction times for their sequenced trials, by the final sequenced block, the two groups had diverged and this was primarily driven by the performance of children with SSD and typical development—not by the participants with CAS. Specifically, the children with SSD who used the uptick pattern tended to *increase* their reaction time at Block 5 relative to Block 1, showing that, for this subset, procedural learning did not occur during this task.

Children with SSD who displayed this pattern may represent a subgroup with a diffuse delayed neural commitment deficit. The Delayed Neural Commitment framework (Nicolson & Fawcett, 2019) has been proposed in children with dyslexia to help explain concurrent delayed language-related skills, phonological impairments, and difficulty automatizing skills in a variety of domains including motor skills. This framework proposes that some children have difficulty building and rebuilding the neural networks that underlie skill acquisition, resulting in these multisystem impairments. Given the multisystem deficits of children who demonstrated this pattern, it is possible that this procedural learning assessment could be used to help screen

children to determine risk for language and motor impairments, two impairments that are often underdiagnosed in the schools, which are associated with academic, social–emotional, and even vocational differences (Cantell et al., 2003; Conti-Ramsden & Durkin, 2012; Durkin & Conti-Ramsden, 2010; Hill & Brown, 2013; Snowling et al., 2001; St Clair et al., 2011). Future work should determine the sensitivity and specificity of procedural learning assessments and the utility of different procedural learning patterns in helping to identify children at risk for communication, reading, and movement disorders. Given that a procedural learning assessment is easily administered via computer or potentially via a mobile application, a parent, teacher’s aide, or classroom volunteer could help to quickly test whole classrooms of children with little training, expense, or effort.

Importantly, use of the uptick pattern was not associated with poorer procedural learning outcomes for those with CAS, whereas it was associated with less (if any) reaction time improvement among children with SSD and typical development. Consequently, this pattern could reflect different causes for the different groups.

Children with CAS are known to exhibit instability and longer articulatory movement durations compared with TD peers during novel word learning tasks (Case & Grigos, 2016). Case and Grigos suggest that these lengthened durations give children with CAS time to process feedback as well as plan and program movements during production of a novel skill. The uptick in reaction time while performing the serial reaction time task may reflect differences in processing, planning, and programming movements at the limb level.

This uptick may also reflect a phenomenon captured by the inverse efficiency score (Townsend & Ashby, 1978). The inverse efficiency score is a metric from cognitive psychology that quantifies the tradeoff between accuracy and reaction time. Accuracy was not measured in our study as children were required to touch the correct square to progress to the next screen. However, an uptick in reaction time could have been associated with the use of increased effort in trying to press more precisely into the center of each box on the screen. The children who demonstrated the uptick pattern (including some children with typical speech and language development) did tend to have lower scores (i.e., at the low end of normal or below) on the Manual Dexterity and Aiming and Catching motor test components, which require precision aiming at targets (e.g., posting coins into a slot, threading a lace through a bead, drawing a path through a trail). Consequently, even though our serial reaction time task is a seemingly easy motor task, the uptick in reaction time may reflect the increased effort children used to compensate for their weaker fine motor abilities (even if those abilities were technically within the normal range as they tended to be in the TD group). Going forward, the inverse efficiency score may be a useful metric of interest during future iterations of this assessment. In addition, other methodologies that assess procedural learning without relying on manual dexterity should also be considered; for instance, the use of eye tracking may be a helpful equalizer.

Children with CAS may have unconsciously attempted the use of different strategies at different times during the task, with some strategies resulting in faster performance than others (Mazzoni & Krakauer, 2006; Taylor & Ivry, 2011). For instance, did faster trials reflect the use of a more effective “ready” position, where the child held their hand in the center of the screen in between trials? This would be a similar strategy to how one stands in a ready position when waiting for the next ball to come over the net in tennis or volleyball. The use of a ready position could help a child to respond faster because (a) it helps the individual to remain focused in between trials and (b) it provides a more favorable location from which to respond, decreasing the proximity between the hand and various response boxes on the screen. While children were not explicitly told to keep their hand in a ready position, some children may have done this naturally, particularly if they had any previous athletic experience where this was trained.

Differences in attention may help to explain the performance of children who produced the uptick pattern and then failed to demonstrate any procedural learning across the sequenced trials. Attention was not measured in the current study, but the extant literature suggests that this is a challenge for half of children with CAS (Lewis et al., 2004; Teverovsky et al., 2009) as well as children with SSD (Beitchman & Inglis, 1991; McGrath et al., 2008). Lewis et al. did a school-age follow-up with 10 children with CAS who were diagnosed at preschool age; of these participants, four of 10 had received an attention-deficit/hyperactivity disorder diagnosis. Similarly, Teverovsky et al. had parents complete a survey on functional issues affecting their child with CAS and found that ~50% of parents reported that focusing and maintaining attention were issues for their child. There is also a high rate of comorbid attention deficit among children with SSD and DLD. Interestingly, some research (Beitchman et al., 1989; Beitchman & Inglis, 1991) show a higher rate of attention deficit among children with linguistic impairments who have resolved SSD rather than persistent SSD. In the current study, participants with SSDs who evidenced the uptick pattern had higher scores on the GFTA-3 but poorer language scores compared to their peers with better procedural learning ability, suggesting that they could possibly be part of a group of children posited to have “general neurodevelopmental immaturity” (Beitchman & Inglis, 1991, p. 107). Beitchman and Inglis characterized children with general neurodevelopmental immaturity as having difficulty acquiring new skills, poorer processing speed, lower attention, poorer language, and *transient* speech deficits. Further procedural learning research on larger cohorts of children with resolved and persistent SSD, with and without attention deficit, is needed to understand the roles of these various mechanisms more fully. In addition, research that matches children based on skillset rather than age may also help to better understand the possibility of a neurodevelopmental immaturity in any of our populations.

It is unknown to what extent attention was a contributing factor to our current findings and whether a more

engaging serial reaction time task would have resulted in a faster rate of learning for our participants with CAS. This is important to consider in the realm of treatment activities. Because we realize that children with CAS require such a high number of practice trials to learn and generalize sequences, treatment activities should be designed to be highly motivating and engaging while limiting distractions.

### *Retention and Consolidation Across Sessions*

The current work tested implicit learning of a simple sequence, and while our participants with CAS demonstrated initial difficulty with this task and required increased exposure to the sequence to learn, they did ultimately demonstrate procedural learning. If we extrapolate these findings to speech treatment that requires a combination of explicit and implicit learning of complex sequences and coordination of multiple articulatory effectors, we can understand why this population may require so many practice trials and treatment sessions to make and retain progress. Future research on larger groups of children with CAS, with a range of language, motor, and cognitive abilities, is needed to fully ascertain procedural learning abilities and patterns in this population.

Importantly, it appeared that, on average, all groups (although not every individual participant) tended to retain knowledge of the sequence from Session 1 to Session 2, which occurred within 4 days of each other. This is critical information because it shows that, even if children with CAS demonstrated slower reaction times and even if they required more trials to initially learn the sequence, they ultimately did show sequence learning and consolidation. This is consistent with the treatment findings demonstrated by Edeal and Gildersleeve-Neumann (2011), which showed learning and generalization of treatment targets in a high-intensity practice condition but not in a low-intensity practice condition. It is unknown to what extent our procedural learning findings are applicable to the learning of a speech sound sequence. This is an empirical question for future consideration.

### *Shared Common Substrate Hypothesis*

Previous literature suggests a global sequencing deficit in a subset of individuals with CAS, such that they have difficulty consistently sequencing speech movements and non-verbal sequences as well (e.g., Nijland et al., 2015; Peter et al., 2018; Shriberg et al., 2012). In the global sequencing deficit framework, motor and cognitive–linguistic deficits are not secondary to CAS, but rather, these deficits, along with the speech deficits, are all due to a shared common substrate. Like Peter et al., we consider the cerebellum a potential region of interest for future investigation as it is central to procedural learning and coordination of motor, visual–spatial, and cognitive–linguistic processes, all areas of difficulty for the participants in our CAS group.

## ***Slower Reaction Time in Children With CAS: Room for Improvement?***

The current findings add to the literature that shows slower reaction time in children with CAS compared to their peers with typical development or other SSDs (Kim et al., 2015). Kim et al. showed that children with CAS evidenced slower immediate and delayed reaction time (30–400 ms, dependent on task) on real-word and pseudo-word tasks. Where some participants with SSD and typical development in the current study seemed to plateau in reaction time improvement during sequenced Blocks 4 and 5, we did not observe this in children with CAS. This finding is consistent with the literature on children with DCD who also evidence slower reaction time on the serial reaction time task but intact procedural learning ability (Wilson et al., 2003). Given that at least half of children with CAS are reported to have fine/gross motor impairments (Gretz, 2013; Iuzzini-Seigel, 2019) and that research (Duchow et al., 2019) reveals that 49% of participants with CAS ( $n = 35$ ) met criteria for a high risk of DCD based on a parent survey, this finding is unsurprising. It is unknown to what extent children with CAS would achieve the same reaction times as their peers with typical development and SSD if given sufficient practice and at what point their reaction time gains would level off—questions that deserve future consideration.

### ***Limitations***

Several limitations were identified in the current study, which provide launch points for future research. Measures of attention were not collected on participants. If children evidenced lower attention during the serial reaction time task, their reaction times may have been negatively impacted by distraction rather than a specific procedural learning difficulty. Given the high rate at which attentional difficulties are known to impact children with CAS (as described above), future research that controls for attentional abilities is needed to better understand the role of this factor on procedural learning performance.

Another limitation was the relative homogeneity of our participants with CAS, who were characterized by a high rate of co-occurring language and motor impairments, which are consistent with more severe reports in the extant literature on children with CAS (e.g., Lewis et al., 2004; Stackhouse & Snowling, 1992; Thoonen et al., 1997) and more severe than other reports (e.g., Case & Grigos, 2016; Murray et al., 2019). Although homogeneity of participant groups is typically considered a study strength, for children with CAS who are known to be heterogeneous (ASHA, 2007) and who vary in language and fine/gross motor abilities, it may reduce the generalizability of our findings.

It should be noted that our groups also differed in nonverbal IQ scores such that children with CAS ( $M = 96$ ) scored significantly lower than the TD group ( $M = 119$ ), who tended to be high scorers, above the average range, on this measure. Despite this and the observed group

differences in language and motor performance, these variables were not included as covariates in our analyses. Dennis et al. (2009) report that covarying for variables such as IQ when studying children with neurodevelopmental diagnoses, such as CAS, is not prudent because these differences are not separable from the population to which these participants belong. If we found that IQ, manual dexterity, or language ability was responsible for all the variance in procedural learning ability, we still could not sort out whether these variables were responsible for procedural learning performance or whether procedural learning ability was driving lower performance on language, motor skills, and IQ. In addition, it is our understanding that the use of IQ as a covariate in neurodevelopmental studies often fails to meet standard assumptions for ANOVA, for instance, that within-group regressions of IQ and our outcome variable will not differ and that residuals will be normally distributed and homoscedastic across groups. Dennis et al. suggest that “previous research on neurocognitive function that used IQ as a matching variable or covariate has produced overcorrected, anomalous, and counterintuitive findings about neurocognitive function” (Dennis et al., 2009, p. 2). Here, we have aimed to avoid this error, but we acknowledge that future work should study procedural learning in large groups of children with CAS across the full IQ, language, and motor ranges to extend and further disambiguate our findings. We had a great deal of overlap between the CAS group and the grammar impaired group, such that it was difficult to fully understand the contribution or relation of each of these diagnoses to procedural learning performance. Future replication should be done with a more diverse sample of participants with grammatical impairments.

We question the extent to which some of the children in the SSD and TD groups may have demonstrated ceiling effects in their procedural learning ability. That is, participants with SSD tended to make more progress during the first session and then showed a less substantial decrease in their reaction time during the second session. In contrast, participants with CAS were significantly slower and demonstrated a large decrease in their reaction time during the second session, more substantial than the change they exhibited during the first session. Future research should test learning of a longer sequence to determine if/when children in different groups achieve maximal speeds during procedural learning training.

Another limitation in the current work is that we only tested learning of a visuospatial sequence. Future work should determine the relation between procedural learning of a visuospatial sequence and response to speech treatment or performance on a word learning/speech learning task (e.g., Case & Grigos, 2016). Studies that control for the gap of days between sessions as well as number of trials per session should be conducted to understand the learning trajectory more fully for children in these populations. What is the predictive value of serial reaction time task performance on a speech sequence training task? If the serial reaction time task can predict the optimal number of trials or schedule of training sessions an individual requires to

learn speech sequences, this would represent a powerful tool for treatment planning. For instance, we could learn if a child requires treatment daily or if there is an acceptable break between treatment sessions during which consolidation can occur without negatively impacting learning and retention. We could also determine if > 100 trials are needed each session or if there is a lower threshold at which learning and retention occur. This is essential for determining the least amount of practice needed to achieve optimal gains—an important efficiency threshold from a public health standpoint.

## Conclusions

The current study found procedural learning differences for children with CAS relative to those with SSD and typical development wherein children with CAS tended to require a greater number of exposures to the sequence to demonstrate learning. Findings provide partial support for the procedural learning deficit hypothesis for children with CAS who evidenced a high rate of co-occurring language and motor deficits as well; procedural learning occurred more slowly in our CAS group, but ultimately, it did occur. It should be noted that participants in our CAS group tended to demonstrate moderate-to-severe language and fine/gross motor deficits, which occurred at a greater rate than is often reported in the extant literature (Case & Grigos, 2016; Murray et al., 2015).

Whereas faster reaction time on each sequenced block was expected, a subset of children in each group evidenced an uptick in reaction time during the second sequenced block. Use of this uptick pattern did not appear to result in a different procedural learning outcome for participants with CAS (compared to those with CAS without this pattern); however, children in the SSD and TD groups who used this pattern tended to demonstrate minimal to no procedural learning gains by the end of the fifth sequenced block and poorer language and motor abilities than their peers who did not demonstrate this pattern. These findings suggest possible divergent causes for this pattern in different populations and may indicate general neurodevelopmental immaturity for a subset of children with SSDs as well. Future research in larger groups of children with CAS, SSD, and typical development with a breadth of fine/gross motor, language, and cognitive abilities is needed to more fully determine procedural learning abilities and patterns in each of these populations.

Future research should also investigate the sensitivity and specificity of this serial reaction time task in identifying children at risk for multisystem speech, language, and motor deficits. Finally, predictive value of performance on the serial reaction time task in relation to treatment outcomes for children with CAS and other communication disorders should be explored.

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