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Jenya luzzini-Seigel

Kristen M. Allison

Ruth Stoeckel

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Tutorial

A Tool for Differential Diagnosis of Childhood Apraxia of Speech and Dysarthria in Children: A Tutorial

Jenya luzzini-Seigel,^a Kristen M. Allison,^b and Ruth Stoeckel^c

^aDepartment of Speech Pathology and Audiology, Marquette University, Milwaukee, WI ^bDepartment of Communication Sciences and Disorders, Northeastern University, Boston, MA ^cDivision of Neurology, Department of Speech Pathology, Mayo Clinic (retired), Rochester, MN

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ABSTRACT

Purpose: While there has been mounting research centered on the diagnosis of childhood apraxia of speech (CAS), little has focused on differentiating CAS from pediatric dysarthria. Because CAS and dysarthria share overlapping speech symptoms and some children have both motor speech disorders, differential diagnosis can be challenging. There is a need for clinical tools that facilitate assessment of both CAS and dysarthria symptoms in children. The goals of this tutorial are to (a) determine confidence levels of clinicians in differentially diagnosing dysarthria and CAS and (b) provide a systematic procedure for differentiating CAS and pediatric dysarthria in children. Method: Evidence related to differential diagnosis of CAS and dysarthria is reviewed. Next, a web-based survey of 359 pediatric speech-language pathologists is used to determine clinical confidence levels in diagnosing CAS and dysarthria. Finally, a checklist of pediatric auditory-perceptual motor speech features is presented along with a procedure to identify CAS and dysarthria in children with suspected motor speech impairments. Case studies illustrate application of this protocol, and treatment implications for complex cases are discussed. **Results:** The majority (60%) of clinician respondents reported low or no confidence in diagnosing dysarthria in children, and 40% reported they tend not to make this diagnosis as a result. Going forward, clinicians can use the feature checklist and protocol in this tutorial to support the differential diagnosis of CAS and dysarthria in clinical practice. Conclusions: Incorporating this diagnostic protocol into clinical practice should help increase confidence and accuracy in diagnosing motor speech disorders in children. Future research should test the sensitivity and specificity of this protocol in a large sample of children with varying speech sound disorders. Graduate programs and continuing education trainings should provide opportunities to practice rating speech features for children with dysarthria and CAS. Supplemental Material: https://doi.org/10.23641/asha.19709146

Childhood apraxia of speech (CAS) and dysarthria are two neurological motor speech disorders that greatly affect intelligibility and response to treatment, but which have divergent bases. CAS is considered a disorder of speech motor planning and programming in the absence of neuromuscular deficits (American Speech-LanguageHearing Association [ASHA], 2007). In contrast, dysarthria is a neuromuscular disorder of motor execution resulting from abnormalities to the strength, range of motion, tone, or precision of movements required for appropriate control of the speech subsystems (i.e., articulatory, respiratory, phonatory, resonatory, and prosodic; Duffy, 2019). Unfortunately, the divergent bases do not result in pathognomonic profiles of speech characteristics, and there is overlap between the speech features that are associated with each disorder. Consequently, differential diagnosis of CAS and dysarthria is challenging.

While many speech pathologists are able to easily list academic descriptions of dysarthria and CAS, in practice,

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Correspondence to Jenya Iuzzini-Seigel: jenya.iuzzini-seigel@ marquette.edu. Publisher Note: This article is part of the Forum: Care of the Whole Child: Key Considerations When Working With Children With Childhood Apraxia of Speech. Disclosure: The authors have declared that no competing financial or nonfinancial interests existed at the time of publication.

a systematic procedure for differentially diagnosing these disorders in children does not exist. In addition, comorbidity of CAS and dysarthria is reported in 4.9% of children with complex neurodevelopmental disorders and occurs over 4 times more frequently among children with certain diagnoses such as Down syndrome (22%; e.g., Wilson et al., 2019). This tutorial will provide a brief overview of the features and procedures that commonly contribute to differential diagnosis of CAS and pediatric dysarthria, report on a survey that examined clinician confidence in diagnosing CAS and dysarthria, and introduce a systematic procedure to support differential diagnosis and treatment planning for these disorders going forward. Operational definitions, procedures, and case studies are included to facilitate learning with the goal of increasing clinical confidence.

Historically, the speech features and procedures used to diagnose CAS and pediatric dysarthria have been based on those reported for adults with acquired apraxia of speech and dysarthria (e.g., Duffy & Josephs, 2012; Rosenbek & Wertz, 1972; Strand et al., 2014; Ziegler, 2002). While adult templates have been useful as a starting place for development of feature lists and procedures, children with these disorders differ from adults in their disorder onset, etiology, and potential for habilitation/ rehabilitation. Furthermore, children with CAS and dysarthria develop speech sounds in the context of their speech motor impairment; thus, their speech presentations are additionally influenced by developmental factors. Consequently, adult systems have limited applicability to children (Morgan & Liégeois, 2010).

In recent years, a growing literature and copious continuing education opportunities have helped to support researchers and clinicians in their differential diagnosis of CAS from other speech sound disorders, an umbrella term that most commonly refers to phonological disorder, articulation disorder, or speech delay (Allison et al., 2020; ASHA, 2007; Iuzzini-Seigel & Murray, 2017; Murray et al., 2015, 2021; Shriberg et al., 2017; Terband et al., 2019). In 2007, the ASHA Ad Hoc Committee on Apraxia of Speech in Children issued a comprehensive technical report that reviewed relevant research on assessment and treatment of CAS. This committee identified three speech features that had gained some consensus in contributing to the differential diagnosis of CAS: (a) inconsistency, (b) lengthened or disrupted coarticulatory transitions, and (c) prosodic disturbance. While the report was incredibly thorough, these features had not yet been validated and specifics on the best way to assess and rate each feature were not included in the report. Since the technical report was released, numerous researchers have attempted to better operationalize and validate these and other features associated with CAS with varying levels of success (Benway & Preston, 2020; Chenausky et al., 2020; Iuzzini-Seigel et al., 2015, 2017; Murray et al.,

2015, 2021; Preston et al., 2021; Shriberg et al., 2017; Terband et al., 2019; Wong et al., 2021). Unfortunately, children with dysarthria have rarely been included in these studies and appear in only small numbers if at all (Iuzzini-Seigel et al., 2017; Maas et al., 2012; Murray et al., 2015). Consequently, it is difficult to know the extent to which these features are evidenced by children with dysarthria as well. Below is a brief review of the most common features associated with diagnosis of CAS and/or dysarthria.

Inconsistency

Inconsistency has long been one of the predominant features used to differentiate CAS from nonmotor-based speech sound disorders (SSDs; e.g., ASHA, 2007; Forrest, 2003; Iuzzini-Seigel et al., 2017; Marquardt et al., 2004), but emerging data suggest that it is less efficacious in differentiating CAS from dysarthria. Inconsistency can be measured at the phonemic (i.e., inconsistency of a speech sound across different words, word-positions, or contexts) or token-to-token (i.e., inconsistency of a word or phrase across multiple repetitions; for a deeper explanation, see the work of Iuzzini-Seigel et al., 2017) levels. Inconsistency in children with CAS can be associated with difficulty consistently planning and programming the appropriate direction, force, timing, and gradation of articulatory movements. Based on the adult literature, individuals with dysarthria may similarly demonstrate inconsistent productions due to variability in their control of vowel durations that signal the postvocalic voicing distinction (Chen, 1970) and in their precision of articulatory contacts due to tongue posturing and timing deficits (Ansel & Kent, 1992). Because speakers with dysarthria often have impairments in multiple speech subsystems (i.e., respiration, phonation, resonance and articulation), task demands can influence speech sound production and result in inconsistency (e.g., consonant devoicing may occur in sentences as the speaker is running out of breath, but not in single words). Consequently, while inconsistency is a fairly sensitive and specific feature to differentially diagnose CAS and nonmotor-based SSDs (Iuzzini-Seigel et al., 2017; Strand & McCauley, 2019), further research is needed to establish the efficacy of this feature in contributing to the differential diagnosis of children with dysarthria and CAS.

Prosodic Disturbance

The efficacy of prosodic disturbance as a differential diagnostic feature of CAS has also been explored by several research groups using various perceptual and acoustic measures (Murray et al., 2015; Shriberg et al., 2011; 2012; 2017; Strand et al., 2013). Prosody, which encompasses suprasegmental features of speech, is a broad domain that

can be assessed with observation of features such as lexical and sentential stress errors, use of equal stress, syllable segmentation/segregation, slow rate, and difficulty with suprasegmental aspects of speech (e.g., pitch inflection, loudness modulation). In addition, acoustic measures such as the pause marker and lexical stress ratio can also be used to determine prosodic disturbance (ASHA, 2007; Chenausky et al., 2020; Fedorenko et al., 2015; Murray et al., 2015; Shriberg et al., 1997, 2017, 2003). The Dynamic Evaluation of Motor Speech Skill (Strand & McCauley, 2019), an instrument designed to assess children with moderate-to-severe CAS, reports 35% sensitivity and 95% specificity for prosodic disturbance in their test and control samples. These data suggest that while this feature may not be present in all children with CAS, if it is present, it is likely to reflect CAS. While this may be true when the comparison group is composed of children with nonmotor-based SSDs, children with dysarthria are also found to have prosodic disturbance (Patel et al., 2012; van Doorn & Sheard, 2001). Whereas prosodic impairment in children with CAS is often associated with flat prosodic contours and staccato-sounding speech, children with dysarthria can have varying prosodic deficits depending on their underlying physiological limitations. Children who have dysarthria secondary to cerebral palsy tend to not only use heightened and exaggerated prosody in an effort to increase intelligibility but also use a more limited set of acoustic cues to mark stress than children with typical development (Patel et al., 2012). Consequently, listeners report that stress is less clear when produced by speakers with cerebral palsy than those with typically developing speech. Slow speaking rate, another feature that occurs among children with CAS, has been found to further disturb prosody for children with dysarthria, particularly for those who are more severely impaired (Darling-White et al., 2018; Patel et al., 2012; Shriberg et al., 2017; Workinger & Kent, 1991).

Difficulty With Coarticulatory Transitions

Another core feature of CAS is difficulty with coarticulatory transitions. Coarticulation occurs when the articulation of different speech segments affects one another, causing an overlap in the articulatory configurations of the different sounds (Terband et al., 2019). Anticipatory coarticulation (i.e., planning for the articulation of a subsequent sound affects the articulation of a preceding sound) and perseveratory coarticulation (i.e., articulatory configuration of one sound affects the articulatory configuration of a subsequent sound) are both normal processes, but the motor planning and programming challenges associated with CAS often result in disruptions of this feature. Difficulty with coarticulatory transitions between sounds, syllables, and words can be observed perceptually in children with CAS as a prolongation of sounds or as a sound addition (e.g., schwa insertion) before or after a word or in between sounds in a cluster (Chenausky et al., 2020; Iuzzini-Seigel et al., 2015; Iuzzini-Seigel & Murray, 2017; Nijland et al., 2002). In addition to these perceptual features of disrupted coarticulation, a number of markers have been documented through acoustic and kinematic analyses, ultrasound, and electropalatography (Nijland et al., 2002; Nittrouer et al., 1989; Noiray et al., 2018; Song et al., 2013; Terband et al., 2019; Timmins et al., 2008; Zharkova et al., 2011, 2012), which are beyond the scope of this tutorial. Although studies of dysarthric adults have reported reduced coarticulation (e.g., Tjaden & Wilding, 2005), prominent disruptions in coarticulatory transitions have not been associated with pediatric dysarthria.

Mayo Features

In addition to the ASHA feature list, there is a checklist of features often referred to as the Mayo checklist (e.g., Shriberg et al., 2011), which includes vowel distortions, difficulty achieving initial articulatory configurations or transitionary movement gestures, equal stress or lexical stress errors, distorted substitutions, syllable segregation, groping, intrusive schwa, voicing errors, slow rate, slow diadochokinetic rates, and increased difficulty with multisyllabic words. The Mayo checklist has been used to support CAS diagnosis in numerous research studies (e.g., Case & Grigos, 2020; Iuzzini-Seigel, 2021; Overby & Caspari, 2015; Overby et al., 2019). Many features in this checklist either are the same as those in the ASHA 3 listed above (i.e., difficulty achieving initial articulatory configurations or transitionary movement gestures, equal stress or lexical stress errors) or can be considered manifestations of those features (e.g., syllable segregation, intrusive schwa, voicing errors, and slow rate can reflect difficulty with coarticulation). Other features such as vowel distortions, distorted substitutions, slow diadochokinetic rates, groping, and increased difficulty with multisyllabic words are unique or consistent with other checklist iterations and include some features that have now been validated in the literature (Chenausky et al., 2020; Davis et al., 1998; Iuzzini-Seigel et al., 2015; Murray et al., 2015; Shriberg et al., 2011). Operational definitions for these features have been published to help support clinical application of this list (Iuzzini-Seigel et al., 2015, 2017; Iuzzini-Seigel & Murray, 2017; Terband et al., 2019).

In prior CAS research, many studies have adopted the criteria that children should display at least four of these CAS features across three different speaking tasks in order to meet criteria for inclusion in CAS groups. However, seven of the features on this Mayo checklist are features that have also been reported as characteristics of pediatric dysarthria. Overlapping features include slow rate, atypical stress, consonant distortions, vowel errors, voicing errors, intrusive schwa, and resonance disturbance (Allison & Hustad, 2018a, 2018b; Haas et al., 2021; Higgins & Hodge, 2002; Hustad et al., 2010; Lee et al., 2014; Workinger & Kent, 1991). Therefore, this checklist is inadequate for differentiating between CAS and dysarthria in children.

Challenges With Differential Diagnosis of Pediatric Dysarthria

While researchers and clinicians have a growing understanding of the best way to diagnose CAS, less is reported about the symptoms and differential diagnostic process for children with dysarthria. In fact, even in research articles about CAS where dysarthria is considered an exclusionary criterion or basis of a second group, the diagnostic criteria for dysarthria are rarely well specified (e.g., Ballard et al., 2010; Iuzzini-Seigel, 2019, 2021; Iuzzini-Seigel et al., 2017; Maas et al., 2019; Murray et al., 2015). Other times, dysarthria may not be mentioned at all despite the possibility of co-occurrence with CAS (Chenuasky et al., 2020; Edeal & Gildersleeve-Neumann, 2011).

Children with dysarthria are reported to have a variety of symptoms that manifest across speech subsystem domains, but the literature often lacks operational definitions for how symptoms should be identified (e.g., Braden et al., 2021; Mei et al., 2020). Consequently, researchers and clinicians have often relied on feature descriptions and motor speech assessments from the adult dysarthria literature (e.g., Duffy, 2019). In recent years, pediatric dysarthria assessments have begun to be developed (Schölderle et al., 2020), and objective measures of intelligibility and acoustic features have been identified that show potential for aiding in dysarthria diagnosis in children (Allison & Hustad, 2018a; Hustad et al., 2015).

Maximum performance tasks (e.g., Rvachew et al., 2005; Thoonen et al., 1999) have also been used to contribute to the diagnosis of dysarthria in children (Allison et al., 2022; Preston et al., 2016, 2017). Tasks including diadochokinesis (DDK; i.e., rapid repeated production of syllables or syllable sequences) and maximum durations of vowels and fricatives are designed to test the speech motor capacities of the respiratory, phonatory, and articulatory systems. While DDK rate and maximum vowel durations have been found to be sensitive and specific at identifying spastic dysarthria in small samples of children with spastic quadriplegia due to cerebral palsy (n = 9; e.g., Thoonenet al., 1996, 1999), to our knowledge, these have not been empirically studied in children with dysarthria due to other etiologies, such as Down syndrome (Kent & Vorperian, 2013; Wilson et al., 2019), who may demonstrate varying profiles of dysarthria features. Given that dysarthria is estimated to occur in ~60% of children with Down syndrome (Wilson et al., 2019) and yet still tends to be underdiagnosed, this represents a big gap in our knowledge. In addition, these protocols may be less directive for speech phenotyping and treatment planning purposes compared to protocols that enable characterization of speech features across multiple speech contexts.

A recent work by Levy et al. (2021) on treatment outcomes in children with dysarthria suggests that diagnostic specifications in the dysarthria literature are becoming more clear. Levy et al. report that their "determination of diagnosis and severity of dysarthria is based on the presence and degree of observable visual characteristics associated with dysarthria (e.g., abnormal orofacial and/or respiratory movement and tone) and audible speech deficits associated with dysarthria (e.g., imprecise articulation, strained vocal quality, decreased articulation rate and vocal intensity, monotone) in at least two of the speech subsystems" (Levy et al., 2021, p. 2303). Levy et al. also report which speech characteristics were demonstrated by each participant in order of "salience," meaning which feature(s) contributed most to a child's percept (Levy et al., 2021). This information may help the reader to understand what each child likely sounded like; although if the reader is not experienced with dysarthric speech, the description may still sound vague.

Other treatment studies on children with dysarthria or with CAS and co-occurring dysarthria report the participants' dysarthria diagnosis and the features demonstrated by each child but do not describe how the dysarthria diagnosis was made (e.g., Fox & Boliek, 2012; Maas et al., 2012; Mei et al., 2020; e.g., How many subsystems needed to be affected to yield a dysarthria diagnosis? What operational definitions were used to guide feature ratings? How many or what features needed to be evident to warrant the dysarthria diagnosis?). Consequently, anecdotal reports suggest that many clinicians may not feel confident in differentially diagnosing motor speech disorders and especially dysarthria as explicit procedures have not yet been agreed upon or made clear in the literature.

Co-occurrence of CAS and Dysarthria in Children

Finally, there is a notable gap in the research literature regarding co-occurrence of CAS and dysarthria in children. Although there is increasing recognition that CAS and dysarthria do co-occur, prior research on CAS has largely excluded children with dysarthria, whereas the dysarthria literature has largely focused on specific populations (e.g., cerebral palsy) and not included CAS comparison groups. A few recent studies have comprehensively described speech presentations of children with specific neurodevelopmental disorders, considering both CAS and dysarthria. Wilson et al. (2019) reported 37.8% incidence of dysarthria, 11% CAS, and 22.2% comorbidity in adolescents with Down syndrome. Likewise, Braden et al. (2021) reported that 100% of their

verbal participants with FOX-P1 mutation (n = 16) demonstrated features of dysarthria and 100% who provided a connected speech sample (n = 14) demonstrated features of apraxia as well. In both studies, dysarthria and CAS diagnoses were reportedly based on clinical judgment of speech features. Although these procedures allowed for detailed characterization of children's speech presentations, they did not explicitly consider features that overlap between the two diagnoses and which can complicate diagnostic determinations. Another complicating factor is that it may be difficult to assess dysarthria based on a paucity of speech resultant from the CAS diagnosis. Likewise, dysarthria may have a greater effect on the child's speech percept with imprecise articulatory contacts, but CAS may have a more significant impact on the child's response to treatment. There is a significant need for clinical guidance that can help speechlanguage pathologists identify CAS and dysarthria in pediatric populations, particularly for children who present with features of both motor speech disorders.

This tutorial will help to fill this gap by (a) using a brief survey study to determine clinician confidence in diagnosing CAS and pediatric dysarthria, (b) providing a list of operationally defined speech features that differentiates those unique to dysarthria or CAS from those that overlap, (c) describing a procedure to support readers in making a diagnosis of CAS and/or dysarthria, and (d) illustrating the use of this diagnostic process through case study examples. A flowchart will also be provided to support the use of this process in clinical practice.

Part I: Survey Study

The goal of this exploratory web-based survey study was to ascertain confidence levels of pediatric speech pathologists in diagnosing CAS and dysarthria. A detailed description of the survey methods including all survey questions is reported in Supplemental Material S1. Speech-language pathologists who had experience working with children were invited to participate. The survey contained six questions pertaining to demographics, five questions about comfort level in differentially diagnosing CAS and dysarthria, and one additional question in which a respondent could provide pertinent open-ended information. Only complete surveys were analyzed. Results will be reported using descriptive statistics.

Results

Demographics

See Supplemental Material S1 for a demographic summary of clinician respondents.

Confidence in Differential Diagnosis of Motor Speech Disorders

Respondents were queried about their confidence in making a differential diagnosis of CAS and dysarthria using a 4-point scale (i.e., not confident, low confidence, moderately confident, and highly confident). Results revealed that while 62% of respondents reported moderate or high confidence in their ability to differentially diagnose CAS, only 40% reported this same level of confidence in diagnosing dysarthria. In contrast, the majority (60%) reported no or low confidence in diagnosing dysarthria. The survey then asked, "To what extent does the following statement describe you? 'I tend to not diagnose children with CAS because I'm not quite sure how to make the diagnosis." to which 20% of respondents reported that this statement mostly or completely describes them. In contrast, 40% of respondents reported that the statement, "I tend not to diagnose dysarthria in children because I'm not quite sure how to make the diagnosis," mostly or completely describes them.

Discussion

Findings from our brief survey of 359 clinician respondents revealed that while the majority of our respondents reported moderate-to-high confidence in diagnosing CAS, the same was not true for dysarthria. Given that these disorders have the same low prevalence of one child/1,000 among children with idiopathic speech delay (Shriberg et al., 2019) and that dysarthria is over 3 times more common than CAS among children with complex neurodevelopmental disorders (Shriberg et al., 2019), limited amount of exposure to children with dysarthria is unlikely to explain the lower level of confidence felt by clinicians in diagnosing dysarthria versus CAS. In addition, both CAS and dysarthria negatively impact intelligibility and can range in severity from mild to severe. For both disorders, severe dysarthria and CAS are often easier to differentiate from other SSDs than in more mild presentations. The last several years have brought increased transparency of diagnostic procedures for CAS in the literature and in continuing education presentations (e.g., diagnostic seminars at state and national conferences, intensive clinical training bootcamps through the Apraxia Kids organization, free trainings provided by the Once Upon a Time Foundation); however, the same transparency and abundance of training opportunities surrounding the dysarthria diagnosis does not yet exist. Pediatric dysarthria research has largely focused on children with cerebral palsy and other specific neurological disorders, and thus there may be less awareness among clinicians of the need to consider dysarthria as a diagnosis in children with neurodevelopmental disorders more broadly. Explicit trainings in which

clinicians become comfortable with operational definitions and gain practice rating features are needed to increase clinical competence and confidence for appropriate diagnosis of dysarthria in children.

Findings that showed that 40% of respondents reported they tend not to diagnose dysarthria because they are not entirely sure how to make the diagnosis are consistent with anecdotal evidence that reports underdiagnosis of dysarthria among children with cerebral palsy and Down syndrome, two populations shown to have an increased rate of dysarthria based on their known neuromuscular deficits (Hustad et al., 2010; Kent et al., 2021; Mei et al., 2014; Sigurdardottir & Vik, 2011). Researchers who investigate the CAS population often tend to exclude children with comorbid dysarthria vet provide only minimal-if any-description about how that diagnostic determination was made. Consequently, little has been written to elucidate the differential diagnostic process for children with dysarthria or with mixed presentations of CAS and dysarthria, further contributing to clinical uncertainty. The following tutorial aims to help fill this gap in knowledge by clearly operationalizing feature definitions and providing case study applications.

Part II: Tutorial

Method

Profile of Childhood Apraxia of speech and Dysarthria Feature List and Rating System

A combined set of auditory-perceptual speech features for rating both dysarthria and CAS characteristics was generated based on prior published literature and current accepted features. For CAS characteristics, the 11 features from the Mayo checklist (Iuzzini-Seigel et al., 2015; Shriberg et al., 2011) were included because this feature set has received the greatest consensus for diagnosis in the current CAS research literature (Allison et al., 2020). For dysarthria characteristics, we compared the full set of deviant auditory-perceptual speech features considered the gold standard for diagnosis of dysarthria in adults (Darley et al., 1969; Duffy, 2019) to feature sets reported in two pediatric studies focused on characterizing auditoryperceptual dysarthria features in children (Schölderle et al., 2020; Workinger & Kent, 1991). Seventeen features that were listed in at least one of the pediatric studies in addition to the full set of adult dysarthria features were included. Similar features with slightly different wording across articles were combined (e.g., "hypernasality" and "nasal emission"; Duffy, 2019), "consistent hypernasality" (Workinger & Kent, 1991), and "hypernasality and nasal emission" (Schölderle et al., 2020) were combined into

"consistent hypernasality" (with or without nasal emission). Importantly, we differentiated "imprecise articulatory contacts" from "consonant distortions" and "vowel errors." Although consonant and vowel distortions are characteristics of both CAS and dysarthria, global lack of precision in articulation of speech sounds is a feature of dysarthria we felt was important to rate separately from segmental errors.

The CAS and dysarthria feature lists were compared for overlap, and six features were found to be associated with both CAS and dysarthria. A master list of features was then compiled, and each feature was coded as being associated with CAS, dysarthria, or both. This initial master list contained 23 features, spanning all speech subsystems (i.e., respiratory, phonatory, resonatory, articulatory, rate/prosodic). Operational definitions for each feature were taken or adapted from published descriptions of the features (Duffy, 2019; Iuzzini-Seigel et al., 2015, 2017).

Because the goal was to generate a feature rating system that could be easily and reliably used in clinical practice, each deviant auditory-perceptual feature was rated on a binary scale (i.e., present or absent) rather than on a Likert scale reflecting severity. Prior research has shown that Likert scale ratings of deviant auditory-perceptual speech features can have poor reliability, particularly in speakers who are impaired across multiple speech dimensions (Allison et al., 2021; Bunton et al., 2007; McHenry, 1999); however, reliability on binary rating scales has been reported to be high in recent research on children with dysarthria (Haas et al., 2021; Schölderle et al., 2020). This master set of features was then piloted by three expert clinicians (the three authors) who rated videos of two children with motor speech impairment due to seizure disorders. Through consensus discussion, some features were combined or omitted if they were either difficult to perceptually rate in children or not necessary for either differential diagnosis or assessing subsystem involvement (11 features). The final consensus list containing 20 features and their operational definitions is shown in Table 1 and is available in Supplemental Material S2 as a template for clinical feature ratings.

This final feature list was used by the three expert raters to judge speech samples from 13 children with epilepsy for another ongoing study (Iuzzini-Seigel et al., 2022). Raters were blinded to any previous speech diagnoses. The presence or absence of each auditory–perceptual speech feature was rated for three video-recorded speech samples from each child: production of single words during administration of the Goldman-Fristoe Test of Articulation–Third Edition (Goldman & Fristoe, 2015), production of words and phrases designed to assess motor speech skills (i.e., build-upon words, challenging multisyllabic words, repetition of the phrase "Buy Bobby a puppy"), and a story retell task. After judging each speech feature, raters each made a final speech diagnosis for the child that could include more than one diagnosis. Feature

Speech subsystem		Feature	Dysarthria/ CAS/both	Operational definition
		· catalo	0,10,0011	
Respiration/phonation	Volume	Low volume or loudness decay	Dysarthria ^a	Consistently quiet voice or voice that gets progressively quieter from beginning to end of utterance
		Excessive loudness	Dysarthria ^a	Consistently loud voice across utterances
		Excess loudness variation	Dysarthria ^a	Voice shows sudden, uncontrolled changes in loudness, sometimes becoming too loud, sometimes too quiet.
	Speech breathing	Effortful/audible inspiration	Dysarthria ^a	Inspiration that is visibly effortful and may also be audible
		Short breath groups	Dysarthria ^a	Phrases are short, possibly because inspirations occur more often than normal. It sounds as if the speaker has run out of air. (note: cannot rate in single-word productions)
	Atypical voice quality		Dysarthria ^a	Voice quality sounds effortful, strained, rough, hoarse, or weak/breathy. Also count if considerable glottal fry is present.
Resonance	Fluctuating resonance/intermittent hyper/ hyponasality		Both ^{a,b}	Resonance intermittently sounds either hyponasal, in which there is not enough airflow out of the nose such that the child sound "stuffy" or hypernasal in which there is too much airflow out of the nose for nonnasal phonemes such as plosives.
	Consistent hypernasality (with or without nasal emission)		Dysarthria ^a	Resonance sounds consistently hypernasal, including weak realization of pressure consonants. Nasal emission may also be present.
Rate/prosody	Slow rate		Both ^{a,b}	Speech rate is atypically slow, including production of syllables, whole words, or phrases. Slowed rate of articulatory movements and/or increased pausing may contribute to reduced rate.
	Atypical stress/reduced stress		Both ^{a,b}	Prosody is characterized by globally reduced or atypical stress. Speech may be monotone/monopitch or have excess-equal stress across syllables.
	Lexical stress errors		CAS ^b	An error in which stress is not placed on the correct syllable in multisyllabic words, either in isolation or embedded in sentences. Stress may be inappropriately equalized across syllables or shifted onto the wrong syllable in the multisyllabic word.
	Syllable segregation		CAS ^b	Brief or lengthy pause between sounds, syllables, or words, such that they are segregated from one another and lacking appropriately smooth transitions. Speech may also be described as having a choppy or staccato-like quality.

 Table 1. ProCAD pediatric motor speech auditory-perceptual feature list.

(table continues)

Table 1. (Continued).

Speech subsystem	Feature	Dysarthria/ CAS/both	Operational definition				
Articulation	Imprecise articulatory contacts	Dysarthria ^a	Overall lack of precision in production of speech sounds, not isolated to specific segments. Speech sounds are distorted due to inadequate sharpness and lack of crisp articulation.				
	Consonant distortions	Both ^{a,b}	A consonant production error in which a speech sound is recognizable as a specific consonant but is not produced accurately (e.g., an /s/ that is produced with lateralization or dentalization)				
	Vowel errors	Both ^{a,b}	A vowel production error in which the vowel is substituted for another vowel or in which the vowel is recognizable as a specific vowel but it is not produced accurately ([aka distorted] e.g., not a prototypical production, may sound like it is in between two vowels).				
	Voicing errors	Both ^{a,b}	A sound is produced as its voicing cognate (e.g., a /p/ that is produced as a /b/). In addition, this could also describe productions that appear to be between voicing categories (i.e., blurring of voicing boundaries)				
	Intrusive schwa	CAS ^b	A schwa is added in between consonants or is inserted at the beginning or end of a word. For example, it may be inserted in between the consonants in a cluster (e.g., /blu/ becomes /bəlu/) or at the end of a word such that /dəg/ becomes /dəgə/.				
	Groping (articulatory searching)	CAS ^b	Silent articulatory searching prior to onset of phonation, possibly in an effort to improve the accuracy of the production (i.e., articulatory groping).				
	Increased difficulty with multisyllabic words	CAS ^b	A disproportionately increased number of errors as the number of syllables increases, as compared to the number of errors on words with fewer syllables				
	Difficulty with initial articulatory configurations and/or transitionary movement gestures	CAS ^b	Initiation of utterance or initial speech sound may be difficult for child to produce and may sound lengthened or uncoordinated. Also, child may evidence lengthened or disrupted coarticulatory gestures or movement transitions from one sound to the next.				

Note. ProCAD = Profile of Childhood Apraxia of speech and Dysarthria; CAS = childhood apraxia of speech.

^aDuffy, 2019. ^bluzzini-Seigel et al., 2015, 2017.

ratings and speech diagnoses were compared between the three judges, and disagreements were resolved through discussions to determine consensus ratings. Interrater reliability of feature ratings was calculated for each child; Randolph's free-marginal multirater kappa statistic (Randolph, 2005) indicated excellent interrater reliability for 11/13 children, and good interrater reliability for the remaining two children. Average percent agreement on the presence of auditory-perceptual features was 92% across all children. These preliminary data suggest strong interrater reliability of the checklist. We were also interested in construct validity, which is the degree to which a test does what it aims to do, in this case, the extent to which our flowchart process accurately identifies CAS and/or dysarthria. Because there is not another tool that has been validated for this purpose, we compared outcomes of the flowchart diagnosis to expert clinical opinion for two different data sets: (a) a population of children with epilepsy with unknown speech diagnosis (n = 13) and (b) a population of children with suspected CAS and developmental coordination disorder but unknown dysarthria status (n = 11). Diagnosis agreed for 77% of participants in the epilepsy data set (Iuzzini-Seigel et al., 2022) and 91% for participants in the CAS + developmental coordination disorder data set (Iuzzini-Seigel et al., 2022). For the purposes of this tutorial, we chose to highlight a few select cases to illustrate and explain how to use this rating system to assess contributions of dysarthria and CAS in children with complex speech presentations.

For the cases highlighted in this tutorial, we determined an atypical auditory-perceptual speech feature to be present if the child exhibited the feature on at least two of the three speaking tasks. Generally, if a feature was present, the child demonstrated the feature multiple times in more than one context. In clinical practice, clinicians should judge whether each auditory-perceptual feature is present on at least two different speaking tasks of varying complexity. Some children may display a feature on all tasks, whereas children with more mild motor speech involvement may not display a feature at the single word level but show it in their connected speech (e.g., low volume or volume decay may only be noticeable in longer utterances). For children with limited verbal output, observation on multiple speech tasks may not be possible. In this case, clinical judgment is needed to determine whether a feature is prominent in the child's speech and should be counted as present.

Using the Profile of Childhood Apraxia of speech and Dysarthria Feature Rating System for Differential Diagnosis

After completing ratings and coming to a consensus on the presence of each auditory-perceptual speech feature, a decision-making flowchart was used to determine whether children had dysarthria, CAS, or both. Please note, this system is designed to help assess the presence of motor speech disorders only. Children may additionally present with other nonmotor-based SSDs. Additional analysis of speech sound error patterns is needed to determine whether a phonological or articulation disorder is present. The flowchart is presented in Figure 1 and summarized as follows:

- 1. Calculate the number of dysarthria-only, CAS-only, and total features the child exhibited. List the subsystem domains in which the child had features present. (See examples in Table 2.)
- 2. Examine dysarthria-only features.
 - a. If NO dysarthria-only features are present, dysarthria can be ruled out.
 - b. If MULTIPLE dysarthria-only features are present across two or more speech subsystem domains, dysarthria can be considered a likely diagnosis. Etiologies of dysarthria in children usually affect the neuromotor system broadly; thus, atypical speech features are most often present in multiple speech subsystem domains.
 - c. If ONLY ONE dysarthria-only feature is present in respiration/phonation or resonance domains, further testing is needed to determine whether dysarthria is present. Isolated features in these domains can have alternate potential causes that need to be considered before a diagnosis can be determined. For example, a hoarse or breathy vocal quality could be due to vocal nodules or another voice disorder, and hypernasality could be due to a submucosal cleft. Hoarse vocal quality and hyponasality are also common in young children with typical development (Schölderle et al., 2020).
 - d. If imprecise articulatory contacts are the ONLY dysarthria-only feature present, dysarthria can be considered a likely diagnosis. Although distortions of individual speech sounds can be associated with CAS or other SSDs, overall weak, slushy, or otherwise globally imprecise articulation that spans across speech segments is a feature unique to dysarthria. Children with imprecise articulatory contacts due to dysarthria usually also exhibit several articulatory features that overlap with CAS (i.e., vowel errors, consonant distortions, and voicing errors) due to their global deficits in execution of articulatory movements.
- 3. Examine CAS-only features.
 - a. If NO CAS-only features are present, CAS can be ruled out.

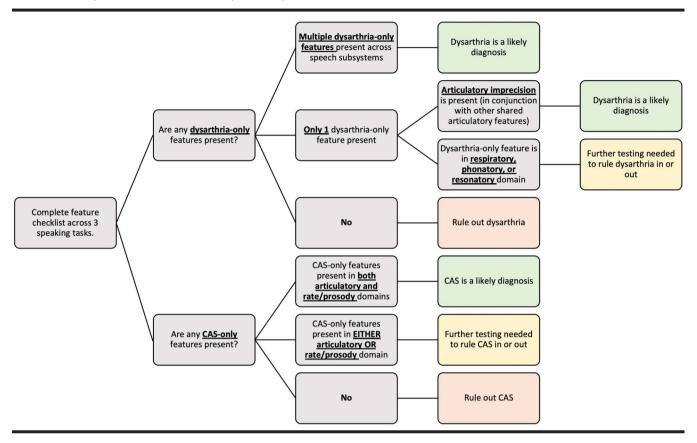


Figure 1. Profile of Childhood Apraxia of speech and Dysarthria flowchart to support the decision-making process for the differential diagnosis of CAS and dysarthria. CAS = childhood apraxia of speech.

- b. If CAS-only features are present in BOTH articulation AND rate/prosody domains, CAS can be considered a likely diagnosis. Core features of CAS relate to disruptions in both articulation and prosody, particularly syllable segmentation and lexical stress. Thus, to be confident in a CAS diagnosis, a child must exhibit features in both of these domains.
- c. If CAS-only features are present in ONLY articulation OR rate/prosody domains, further assessment or analysis is needed to determine whether CAS is present. Quantitative analysis of inconsistency and detailed analysis of segmental error patterns may be helpful in confirming or ruling out CAS in these cases. Isolated CAS-only features may be present in children with a history of CAS who now only present with residual symptoms or children with speech motor delay.

Case Studies

The Profile of Childhood Apraxia of speech and Dysarthria (ProCAD) diagnostic features framework was applied to four children for whom video-recorded speech sample data from other studies were available. Consensus ratings of features for each child are presented in Table 2. The decision-making flowchart described above was used to examine how these features contributed to an understanding of each child's communication impairment and consideration of how intervention would be influenced by identification of specific features or groups of features. General goals for speech are offered for each case, with the acknowledgement that additional support for other areas of impairment will most likely also need to be included in a treatment plan, such as consideration for augmentative and alternative communication, specific goals for language intervention and support for literacy.

Child 1

Child 1 is a girl who was age 4;7 (years;months) at the time of evaluation. She has a diagnosis of cerebral palsy, seizure disorder, and feeding difficulties. Language testing indicated delays in both receptive and expressive language. Child 1 spoke primarily in single words and short phrases.

Features of Child 1's speech were notable across multiple subsystems. See Supplemental Material S3 for a video example of this child's speech. These included low volume, atypical voice quality, fluctuating resonance, and

Speech subsystem	Speech feature		Dysarthria/ CAS/both	Child 1	Child 2	Child 3	Child 4
Respiration/phonation (R/P)	Volume	Low volume or loudness decay Excessive loudness	Dysarthria ^a Dysarthria ^a	X			
	Speech breathing	Excess loudness variation Effortful/audible inspiration Short breath groups	Dysarthria ^a Dysarthria ^a Dysarthria ^a	х			
Resonance (Res)	Atypical voice quality fluctuating resonance/	Dysarthria ^a Both ^{a,b} Dysarthria ^a	X X	Х	Х		
Rate/prosody (Pros)	consistent hypernasality (with or without nasal emission) Slow rate Atypical stress/reduced stress Lexical stress errors Syllable segregation imprecise articulatory contacts consonant distortions vowel errors voicing errors intrusive schwa groping (articulatory searching) increased difficulty with multisyllabic words difficulty with initial artic configs/transitionary movement		Both ^{a,b} Both ^{a,b}	Х	х	Х	х
Articulation (Artic)			CAS ^b CAS ^b Dysarthria ^a Both ^{a,b} Both ^{a,b}	Х	х	X X X	x x
Aniculation (Anic)				X X	X X	X X	X X
			Both ^{a,b} CAS ^b CAS ^b	Х	Х	Х	х
			CAS ^b CAS ^b		х	Х	X X
gestures No. of Dys-only features No. of CAS-only features				3 0	1 2	1 3	0
No. of total features Subsystems involved				9 ⊠ R/P	7 ⊠ R/P	9 □ R/P	8 □ R/P
				⊠ Res ⊠ Pros ⊠ Artic	□ Res ⊠ Pros ⊠ Artic	⊠ Res ⊠ Pros ⊠ Artic	□ Res ⊠ Pros ⊠ Artic
Severity rating Diagnosis				Severe Dysarthria	Profound CAS	Severe/profound CAS + dysarthria	Moderate CAS

 Table 2. Consensus auditory-perceptual feature ratings for case studies.

Note. CAS = childhood apraxia of; Dys = dysarthria.

^aDuffy, 2019. ^bluzzini-Seigel et al., 2017, or luzzini-Seigel & Murray, 2017.

imprecise articulatory contacts. Intelligibility was judged to be severely reduced in connected speech. She was rated as having three features consistent with dysarthria and five features overlapping with dysarthria and CAS. Referring to the decision tree for this child (see Figure 2), there are multiple features of dysarthria across all five speech subsystems and no features unique to CAS, leading to a diagnosis of dysarthria. While CAS does not appear to have a significant contribution to Child 1's speech impairment at this time, given her limited verbal skill, it would be reasonable to revisit the features that are present in her speech as verbal skills increase.

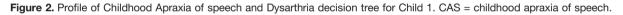
Given Child 1's presentation, intervention would involve a subsystems approach that might initially prioritize improved respiratory support and coordination of respiration and phonation to establish a foundation for improving verbal skills (Pennington et al., 2010, 2013). The child would practice coordinating the onset of phonation with exhalation through production of prolonged vowels and then move to syllables and syllable sequences (short functional phrases). Practice is organized according to principles of motor learning, such as frequent practice, manipulation of blocked versus random practice, and type and frequency of feedback to move from acquisition to retention and generalization. The severity of Child 1's communication disorder and her developmental level influence her readiness for an approach requiring greater verbal ability and self-monitoring, such as the Speech Intelligibility Treatment (Levy et al., 2021).

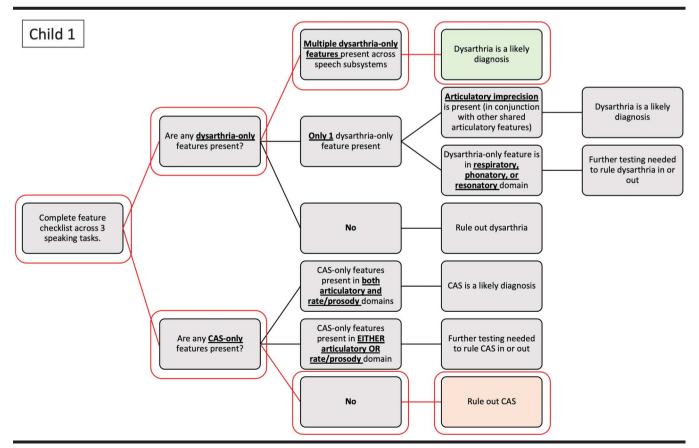
Child 2

Child 2 is a girl evaluated at age 6;9. She has a history of seizures during sleep, for which she takes multiple medications. Genetic testing was normal. Child 2 has a prior diagnosis of CAS and has a cousin with CAS.

At the time of assessment, Child 2 was speaking in single words. Standardized language testing indicated mildly delayed receptive language and moderately delayed expressive language.

Features of Child 2's speech included atypical/ strained voice quality, slow rate, syllable segregation, and difficulty with initial articulatory configurations. Ratings included one feature specific to dysarthria, two features specific to CAS, and four overlapping features. Based on ratings of CAS-only features in both articulatory and rate/ prosody domains (see Figure 3), a primary diagnosis of CAS was considered likely. The feature of voice quality suggests the need to rule out a possible later diagnosis of





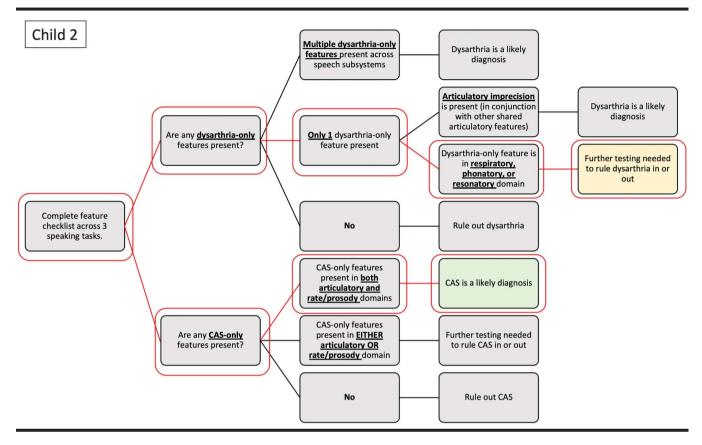


Figure 3. Profile of Childhood Apraxia of speech and Dysarthria decision tree for Child 2. CAS = childhood apraxia of speech.

dysarthria when verbal skills have increased and the respiratory and phonatory systems can be more fully assessed.

For Child 2, syllable segregation had a significant effect on intelligibility as noted by all three expert raters. This child also had noticeable difficulty with initial articulatory configurations and sequencing movement gestures. Intervention for this child might initially focus on accuracy of articulatory movement gestures and attention to smooth coarticulation in syllables and syllable sequences. An example of a framework that involves practice in a hierarchy of complexity with varying levels of cueing is dynamic temporal and tactile cueing (DTTC; Strand, 2020). Adapting goals to incorporate elements of treatment for dysarthria and addressing respiration/phonation and rate (Pennington et al., 2013) could also be used to evaluate Child 2's response and further inform diagnosis.

Child 3

Child 3 is a boy who was age 5;7 at the time of evaluation. He had been referred for genetic testing due to concerns that he could not cough, laugh, or sneeze. Genetic testing revealed a mutation on FOXP2. Medical history also includes removal of adenoids and placement of ventilating ear tubes.

Language testing indicated delays in both receptive and expressive language. Spontaneous language was

characterized by utterances of moderately reduced length and complexity relative to age expectations.

Child 3's speech included fluctuating resonance, atypical stress with syllable segregation, increased difficulty with multisyllabic words, and imprecise articulatory contacts. In addition to the features captured on this measure, he exhibited several identifiable error patterns.

Ratings for Child 3 showed three features specific to CAS, five features overlapping with dysarthria and CAS, and one feature specific to dysarthria. This combination of feature ratings suggested a mixed disorder. CAS was considered to be predominant based on pervasiveness of syllable segregation, with dysarthria suggested by imprecise articulatory contacts (see Figure 4). While not captured on this feature rating tool, further evaluation for error patterns that were noted and the possible contribution of phonological impairment would be appropriate.

For a child with a mixed diagnosis, attention to the affected subsystems is important for informing treatment decisions. Child 3 was rated as having adequate respiratory/ phonatory support; therefore, the focus of intervention would be on increasing articulatory accuracy, to address imprecision that might be due to mild weakness as well as inaccuracies due to the motor planning/programming impairment. A motor-based framework such as DTTC

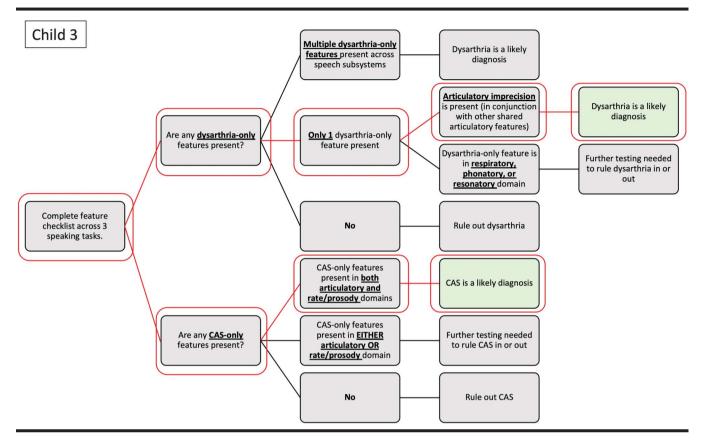


Figure 4. Profile of Childhood Apraxia of speech and Dysarthria decision tree for Child 3. CAS = childhood apraxia of speech.

(Strand, 2020) could be adapted to incorporate specific work on identified phonological patterns as well. Consideration should be given to addressing the prosodic disturbance in a framework such as Rapid Syllable Transition (e.g., McCabe et al., 2020) or Treatment for Establishing Motor Program Organization (Miller et al., 2021) given that this child was able to speak at phrase level. These prosody-based approaches use nonsense words, presuming that practice of these sequences without the ability to rely on existing motor plans allows the child to focus mostly on suprasegmental aspects of production.

Child 4

Child 4 is a boy who was 4 years of age at the time of evaluation. It was reported that he had difficulty with latch and suck as an infant and would often choke or projectile vomit during and after feeding, continuing until age 3 years. He additionally had a reported history of tonsillectomy and adenoidectomy, frenulectomy, and pressure equalization tube insertion. Child 4 was said to be understood by parents, siblings, unfamiliar listeners, and peers less than 50% of the time and used a dedicated augmentative and alternative communication device to supplement his limited verbal output. Both receptive and expressive language skills were judged to be delayed. Spontaneous verbal output was limited. Child 4's speech production was characterized by vowel errors, consonant distortions, voicing errors, difficulty with initial articulatory configurations/transitionary movement gestures, intrusive schwa, syllable segregation, and lexical stress errors. Child 4 was rated as having four features specific to CAS and four overlapping features for CAS and dysarthria. The affected subsystems were articulation and rate/prosody. The decision tree leads to a diagnosis of CAS (see Figure 5).

In this case, intervention would prioritize motor planning and programming for speech production using principles of motor learning to work on improving accuracy of production of syllables and syllable sequences within a hierarchy of levels of cueing and linguistic complexity. Given his limited verbal output, a framework intended for younger or more severe children such as DTTC (Strand, 2020) should be considered. Additional case studies are included in Supplemental Material S4 for practice purposes.

Implementation: Potential Barriers and Potential Solutions

The current tutorial provided a list of speech features and procedures to support readers in making a diagnosis of

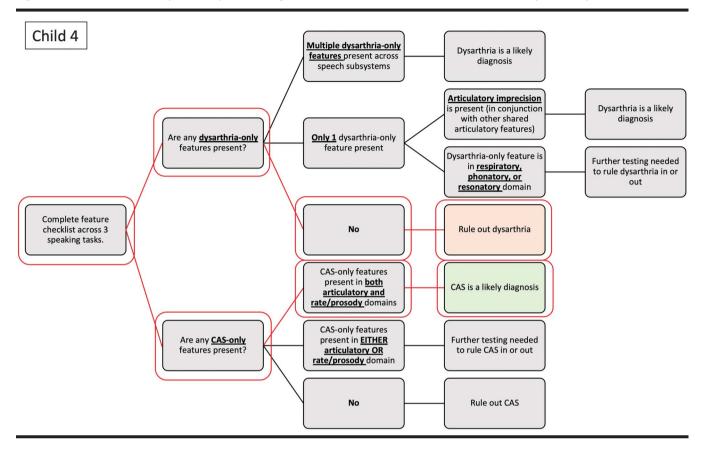


Figure 5. Profile of Childhood Apraxia of speech and Dysarthria decision tree for Child 4. CAS = childhood apraxia of speech.

CAS and/or dysarthria and to illustrate the use of this diagnostic process through case study examples. While the case studies sampled four distinct types of challenging clinical cases, there are additional clinical presentations that may be difficult to assess using the proposed procedure. Given the number of overlapping features, there may be instances when it is difficult to disambiguate CAS from dysarthria if the more discriminative features are not exhibited consistently across tasks. For children with verbal output limited to single syllables or monosyllable words, some features (e.g., syllable segregation) may not be observable and therefore cannot be adequately assessed. In such cases, we recommend marking the characteristic as n/a in the chart. As children begin producing more speech through therapy and/or development, it is possible that features may emerge that were not previously observed. For example, presence of short phrases/breath groups cannot be assessed unless a child is producing connected speech. Thus deficits in breath support for speech may become apparent later, at which point a dysarthria diagnosis may be considered. In these cases, we recommend confirming the presence/ absence of any features that can be observed and indicate that CAS/dysarthria cannot be ruled out or is suspected based on features that are demonstrated.

Comorbid language impairment is extremely common in children with motor speech disorders and can also impact a child's speech presentation. When speaking tasks exceed a child's language ability or are linguistically challenging (e.g., nonword repetition, sentence repetition, or story retell), atypical speech features may not reflect a motor speech etiology. For instance, some children may demonstrate low vocal intensity or inconsistency when performing tasks with high language demands. Iuzzini-Seigel et al. (2017) showed that while inconsistency on repetitions of "Buy Bobby a puppy" could differentiate CAS from a phonological disorder, children with developmental language disorder were also inconsistent in their production of this phrase. Prior research has also shown that children with language impairments show disproportionate increases in articulatory variability in tasks with increased language demands, compared to children with typical language skills (Vuolo & Goffman, 2018). Because of this interaction between the speech and language systems, it is essential to formally assess each child's language abilities and to consider the potential impact of their language abilities on their speech output.

Children who have undergone copious amounts of treatment may also demonstrate variable output across

tasks. For instance, a child with residual CAS may not exhibit errors in simple contexts (e.g., single word level) but may evidence breakdowns in complex tasks (e.g., repetition of "Buy Bobby a puppy," sentence repetition, trisyllable DDK). Consequently, treatment history can also contribute essential information to the assessment process and help to guide treatment.

Why Were Features That Overlap Between CAS and Dysarthria Included in the Checklist?

Although several auditory-perceptual speech features overlap between CAS and dysarthria and therefore may not enhance diagnostic accuracy, they remain important for treatment planning. It is helpful to thoroughly characterize speech features across subsystem domains in order to understand each child's speech profile and identify contributing factors to intelligibility deficits. Identifying which speech subsystems are affected has important implications for treatment decision making (Hodge & Wellman, 1999). Importantly, overlapping features may be attributed to either CAS or dysarthria, but if both diagnoses are present, overlapping features may not be able to be clearly attributed to one diagnosis or the other. In the case of a child with both dysarthria and CAS, dynamic assessment can be helpful to determine how children respond to different strategies and whether a CAS or dysarthria treatment approach may be most helpful.

Why Were Some Common CAS and Dysarthria Features (e.g., Subtypes of Atypical Voice Quality and Inconsistency) Excluded From the Checklist?

The purpose of this checklist is to help clinicians to determine whether dysarthria and/or CAS is present and to assess which speech subsystems are impacted. Therefore, we included key features within each subsystem domain but omitted or combined some more granular dysarthria features that are challenging to reliably judge at a perceptual level (Allison et al., 2020; Bunton et al., 2007; McHenry, 1999). For example, in the respiratory/phonatory domain, we condensed related features into single categories to make them easier to judge (e.g., breathy, rough/hoarse, and strained-to-strangled voice quality would all be counted as "atypical voice quality"). Similarly, in the rate/ prosody domain, monopitch, monoloudness, and excess and equal stress would all be counted as "atypical/reduced stress." These more specific features can be important for dysarthria treatment planning, as different intervention approaches may be needed for children with predominant spasticity and vocal hyperfunction (e.g., strained-to-strangled voice quality) compared to children with predominant weakness and vocal hypofunction (e.g., breathy voice quality; Duffy, 2019). However, for the purposes of differentiating dysarthria from CAS and assessing subsystem involvement, the included broader features should be adequate. If respiratory/phonatory or resonatory features are identified as a problem area, additional in-depth assessment may be needed to further evaluate the child's speech characteristics, including more in-depth auditory– perceptual assessment, instrumental evaluation (e.g., nasendoscopy), or acoustic analysis.

While inconsistency is considered a sensitive and specific feature for supporting the differential diagnosis of CAS and nonmotor-based SSDs (e.g., Iuzzini-Seigel et al., 2017; Strand & McCauley, 2019), there is inadequate evidence at this time that it supports differential diagnosis between CAS and dysarthria. In addition, and perhaps more importantly, whereas the other features that are included on the checklist may be judged at the perceptual level across a variety of tasks, inconsistency measures often require administration of a particular assessment, some amount of data processing, and/or calculation of a metric to determine if the child should be considered inconsistent or not. Consequently, we opted to leave this important feature off of the current checklist but recommend that it is included if the clinical intent is to differentiate CAS from nonmotor-based SSDs

If Multiple Disorders Are Co-occurring, How Do I Determine Which Is the Primary Disorder?

In some children, a CAS- or dysarthria-only symptom may drive the general percept of the child's speech. For instance, in a child who has overwhelmingly imprecise articulatory contacts or short breath groups and labored respiration, we may say that the child has a primary contribution of dysarthria even if that child also demonstrates features that are CAS only. Other times, a short trial period of therapy may reveal which disorder is driving the child's response in treatment. For instance, Child 3 participated in a trial of DTTC treatment. Although he did demonstrate pervasive imprecise articulatory contacts throughout our speech assessment, during the treatment trial, it was clear that CAS was creating the greatest barrier to his remediation. Consequently, we want to consider the contribution of each diagnosis to a child's intelligibility, but also the impact of each diagnosis on remediation. A child who has a history of CAS may eventually demonstrate error patterns reflective of a phonological impairment. Although it may be appropriate to select phonological treatment targets at that time, we would still likely need to train these targets while applying the principles of motor learning to continue to address the underlying CAS diagnosis.

Am I Likely to See a Child With CAS or Dysarthria Who Has a Co-occurring Articulation or Phonological Disorder?

Having a motor speech disorder does not preclude an articulation or phonological disorder. In fact, children

with neurodevelopmental disorders often have co-occurring impairments in motor speech, language, and cognitive skills, all of which can increase the likelihood of phonological delays or disorders (Shriberg et al., 2019). Even among children with CAS who are largely inconsistent, we often see phonological patterns or articulation errors start to emerge, especially as the child starts to respond to treatment (e.g., Crosbie et al., 2005; Dodd & Bradford, 2000; Iuzzini & Forrest, 2010). Additional in-depth phonological analysis may be needed to fully characterize children's speech sound error patterns, and patterns should be interpreted in the context of the child's motor speech constraints. For example, many children with dysarthria and CAS have trouble regulating voicing and tend to produce predominantly voiced consonants (e.g., Iuzzini-Seigel et al., 2015; Lewis et al., 2004). Based on a phonological process analysis, this pattern may be described as prevocalic voicing; however, if this pattern is due to a child's motor speech constraints, it will likely require a motor-based treatment approach to correct (i.e., establishing better control of voicing onset/offset) rather than a phonological approach (e.g., through auditory discrimination/minimal pairs). When treating a child with a motor speech disorder who has a co-occurring phonological or articulation disorder, principles of motor learning should be used to guide treatment and promote the greatest gains, but targets may be selected with phonological or articulation goals in mind.

Can the ProCAD Checklist and Procedure Be Used for Re-evaluation As Well?

The original intent of this checklist protocol was for diagnostic and treatment planning purposes; it can, however, be used to monitor changes to the speech profile over time as well. While features may change, emerge, or remediate with treatment, a reduction in features should not suggest that the child has been "cured" of CAS or dysarthria as these are lifelong, neurological disorders. Rather, a change or reduction in features can help to drive revision of goals. Likewise, at initial evaluation, a child may not provide an adequate speech sample such that CAS or dysarthria can neither be confirmed nor ruled out. In such cases, re-evaluation once the child is able to produce a more substantive speech sample may allow for confirmation/or ruling out of one or both diagnoses.

Conclusions

CAS and dysarthria are low-incidence disorders, with prevalence of each estimated at one child per 1,000 (Shriberg et al., 2019). However, the presence of these disorders can have a significant impact on a child's ability to communicate, particularly for children with comorbid diagnoses. Despite the frequency of complex motor speech

presentations, particularly in children with neurodevelopmental disorders, tools to help guide clinical pediatric motor speech assessment are lacking. A growing research base and numerous continuing education opportunities focused on differential diagnosis of CAS have emerged in recent years, but less information is available pertaining to the diagnosis of pediatric dysarthria. Consequently, clinician respondents to our survey reported lower confidence levels in diagnosing dysarthria than CAS and 40% reported that they sometimes fail to diagnose dysarthria because they are not sure of how that diagnosis should be made. The current tutorial provides a procedure to support the identification and differential diagnosis of CAS and dysarthria, helping to fill a knowledge gap in the field. The flowchart was developed to assist clinicians in applying the checklist of perceptual CAS and dysarthria features to the rating of single word and connected speech samples. This procedure can help determine whether each of these disorders should be ruled in or out, identify speech subsystems that should be addressed in treatment, and provide direction for additional in-depth speech assessment if needed.

As this checklist was newly developed as part of an ongoing research study, its diagnostic accuracy has not yet been established in a large cohort of children with and without motor speech disorders, across the age span, and with and without complex medical profiles. In addition, to date, we have only applied this checklist to native English speakers, and consequently, it is unknown to what extent this tool will be efficacious for non-English speakers or multilingual speakers of languages that are not stress timed. Preliminary estimates of construct validity range between 77% and 91% for diagnostic agreement between our flowchart and expert clinicians for children with epilepsy and those with suspected CAS and developmental coordination disorder. As such, this tool should be viewed as a clinical framework for guiding motor speech assessment in children. Future research should focus on establishing the sensitivity and specificity of this procedure in a large cohort of children with varying speech profiles and on comparing the diagnostic accuracy of these auditory perceptual features to acoustic and instrumental measures. Future work will also determine the effectiveness of this procedure in clinical practice among clinicians with varying specializations and levels of experience. It is essential that more continuing education opportunities are developed to help clinicians continue to gain confidence and expertise diagnosing motor speech disorders thereby increasing the quality of care for children with these challenging diagnoses.

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References

- Allison, K. M., Cordella, C., Iuzzini-Seigel, J., & Green, J. R. (2020). Differential diagnosis of apraxia of speech in children and adults: A scoping review. *Journal of Speech, Language, and Hearing Research, 63*(9), 2952–2994. https://doi.org/10. 1044/2020_JSLHR-20-00061
- Allison, K. M., & Hustad, K. C. (2018a). Acoustic predictors of pediatric dysarthria in cerebral palsy. *Journal of Speech, Lan*guage, and Hearing Research, 61(3), 462–478. https://doi.org/ 10.1044/2017_JSLHR-S-16-0414
- Allison, K. M., & Hustad, K. C. (2018b). Data-driven classification of dysarthria profiles in children with cerebral palsy. *Journal of Speech, Language, and Hearing Research, 61*(12), 2837–2853. https://doi.org/10.1044/2018_JSLHR-S-17-0356
- Allison, K. M., Nip, I., & Rong, P. (2022). Utility of automated kinematic diadochokinesis analysis for identification of dysarthria in children [Manuscript submitted for publication].
- Allison, K. M., Russell, M., & Hustad, K. C. (2021). Reliability of perceptual judgments of phonetic accuracy and hypernasality among speech-language pathologists for children with dysarthria. *American Journal of Speech-Language Pathology*, 30(3S), 1558–1571. https://doi.org/10.1044/2020_AJSLP-20-00144
- American Speech-Language-Hearing Association. (2007). Childhood apraxia of speech [Technical report]. https://doi.org/10.1044/ policy.TR2007-00278
- Ansel, B. M., & Kent, R. D. (1992). Acoustic–phonetic contrasts and intelligibility in the dysarthria associated with mixed cerebral palsy. *Journal of Speech and Hearing Research*, 35(2), 296–308. https://doi.org/10.1044/jshr.3502.296
- Ballard, K. J., Robin, D. A., McCabe, P., & McDonald, J. (2010). A treatment for dysprosody in childhood apraxia of speech. *Journal of Speech, Language, and Hearing Research, 53*(5), 1227–1245. https://doi.org/10.1044/1092-4388(2010/09-0130)
- Benway, N. R., & Preston, J. L. (2020). Differences between school-age children with apraxia of speech and other speech sound disorders on multisyllable repetition. *Perspectives of the ASHA Special Interest Groups*, 5(4), 794–808. https://doi.org/ 10.1044/2020_PERSP-19-00086
- Braden, R. O., Amor, D. J., Fisher, S. E., Mei, C., Myers, C. T., Mefford, H., Gill, D., Srivastava, S., Swanson, L. C., Goel, H., Scheffer, I. E., & Morgan, A. T. (2021). Severe speech impairment is a distinguishing feature of FOXP1-related disorder. *Developmental Medicine & Child Neurology*, 1–10. https://doi.org/10.1111/dmcn.14955
- Bunton, K., Kent, R. D., Duffy, J. R., Rosenbek, J. C., & Kent, J. F. (2007). Listener agreement for auditory–perceptual ratings of dysarthria. *Journal of Speech, Language, and Hearing Research*, 50(6), 1481–1495. https://doi.org/10.1044/1092-4388(2007/102)
- Case, J., & Grigos, M. I. (2020). A framework of motoric complexity: An investigation in children with typical and impaired

speech development. Journal of Speech, Language, and Hearing Research, 63(10), 3326–3348. https://doi.org/10.1044/2020_ JSLHR-20-00020

- Chen, M. (1970). Vowel length variation as a function of the voicing of the consonant environment. *Phonetica*, 22(3), 129–159. https:// doi.org/10.1159/000259312
- Chenausky, K. V., Brignell, A., Morgan, A., Gagné, D., Norton, A., Tager-Flusberg, H., Schlaug, G., Shield, A., & Green, J. R. (2020). Factor analysis of signs of childhood apraxia of speech. *Journal of Communication Disorders*, 87, 106033. https://doi.org/ 10.1016/j.jcomdis.2020.106033
- Crosbie, S., Holm, A., & Dodd, B. (2005). Intervention for children with severe speech disorder: A comparison of two approaches. *International Journal of Language & Communication Disorders*, 40(4), 467–491. https://doi.org/10.1080/13682820500126049
- Darley, F. L., Aronson, A. E., & Brown, J. R. (1969). Differential diagnostic patterns of dysarthria. *Journal of Speech and Hearing Research*, 12(2), 246–269. https://doi.org/10.1044/jshr. 1202.246
- Darling-White, M., Sakash, A., & Hustad, K. C. (2018). Characteristics of speech rate in children with cerebral palsy: A longitudinal study. *Journal of Speech, Language, and Hearing Research*, 61(10), 2502–2515. https://doi.org/10.1044/2018_JSLHR-S-17-0003
- Davis, B. L., Jakielski, K. J., & Marquardt, T. P. (1998). Developmental apraxia of speech: Determiners of differential diagnosis. *Clinical Linguistics & Phonetics*, 12(1), 25–45. https:// doi.org/10.3109/02699209808985211
- Dodd, B., & Bradford, A. (2000). A comparison of three therapy methods for children with different types of developmental phonological disorder. *International Journal of Language & Communication Disorders*, 35(2), 189–209. https://doi.org/10. 1080/136828200247142
- **Duffy, J. R.** (2019). Motor speech disorders e-book: Substrates, differential diagnosis, and management. Elsevier Health Sciences.
- Duffy, J. R., & Josephs, K. A. (2012). The diagnosis and understanding of apraxia of speech: Why including neurodegenerative etiologies may be important. *Journal of Speech, Language, and Hearing Research*, 55(5), S1518–S1522. https://doi. org/10.1044/1092-4388(2012/11-0309)
- Edeal, D. M., & Gildersleeve-Neumann, C. E. (2011). The importance of production frequency in therapy for childhood apraxia of speech. *American Journal of Speech-Language Pathology*, 20(2), 95–110. https://doi.org/10.1044/1058-0360(2011/09-0005)
- Fedorenko, E., Hsieh, P. J., & Balewski, Z. (2015). A possible functional localizer for identifying brain regions sensitive to sentence-level prosody. *Language, Cognition and Neuroscience,* 30(1–2), 120–148. https://doi.org/10.1080/01690965.2013.861917
- Forrest, K. (2003). Diagnostic criteria of developmental apraxia of speech used by clinical speech-language pathologists. *Ameri*can Journal of Speech-Language Pathology, 12(3), 376–380. https://doi.org/10.1044/1058-0360(2003/083)
- Fox, C. M., & Boliek, C. A. (2012). Intensive voice treatment (LSVT LOUD) for children with spastic cerebral palsy and dysarthria. *Journal of Speech, Language, and Hearing Research*, 55(3), 930–945. https://doi.org/10.1044/1092-4388(2011/10-0235)
- Goldman, R., & Fristoe, M. (2015). Goldman-Fristoe Test of Articulation-Third Edition [Assessment instrument]. Pearson Assessments.
- Haas, E., Ziegler, W., & Schölderle, T. (2021). Developmental courses in childhood dysarthria: Longitudinal analyses of auditory-perceptual parameters. *Journal of Speech, Language, and Hearing Research, 64*(5), 1421–1435. https://doi.org/10. 1044/2020_JSLHR-20-00492

- Higgins, C., & Hodge, M. (2002). Vowel area and intelligibility in children with and without dysarthria. *Journal of Medical Speech-Language Pathology*, 10, 271–277.
- Hodge, M. M., & Wellman, L. (1999). Management of children with dysarthria. In A. Caruso & E. Strand (Eds.), *Clinical management of motor speech disorders in children* (pp. 209–280). Thieme.
- Hustad, K. C., Gorton, K., & Lee, J. (2010). Classification of speech and language profiles in 4-year-old children with cerebral palsy: A prospective preliminary study. *Journal of Speech, Language, and Hearing Research, 53*(6), 1496–1513. https://doi.org/10.1044/1092-4388(2010/09-0176)
- Hustad, K. C., Oakes, A., & Allison, K. (2015). Variability and diagnostic accuracy of speech intelligibility scores in children. *Journal of Speech, Language, and Hearing Research*, 58(6), 1695–1707. https://doi.org/10.1044/2015_JSLHR-S-14-0365
- Iuzzini, J., & Forrest, K. (2010). Evaluation of a combined treatment approach for childhood apraxia of speech. *Clinical Linguistics & Phonetics*, 24(4–5), 335–345. https://doi.org/10.3109/ 02699200903581083
- Iuzzini-Seigel, J. (2019). Motor performance in children with childhood apraxia of speech and speech sound disorders. *Journal* of Speech, Language, and Hearing Research, 62(9), 3220–3233. https://doi.org/10.1044/2019_JSLHR-S-18-0380
- Iuzzini-Seigel, J. (2021). Procedural learning, grammar, and motor skills in children with childhood apraxia of speech, speech sound disorder, and typically developing speech. *Journal of Speech, Language, and Hearing Research, 64*(4), 1081–1103. https://doi. org/10.1044/2020_JSLHR-20-00581
- Iuzzini-Seigel, J., Allison, K., & Stoeckel, R. (2022). Motor speech diagnoses in children with epilepsy [Manuscript in preparation].
- Iuzzini-Seigel, J., Hogan, T. P., & Green, J. R. (2017). Speech inconsistency in children with childhood apraxia of speech, language impairment, and speech delay: Depends on the stimuli. *Journal of Speech, Language, and Hearing Research, 60*(5), 1194–1210. https://doi.org/10.1044/2016_JSLHR-S-15-0184
- Iuzzini-Seigel, J., Hogan, T. P., Guarino, A. J., & Green, J. R. (2015). Reliance on auditory feedback in children with childhood apraxia of speech. *Journal of Communication Disorders*, 54, 32–42. https://doi.org/10.1016/j.jcomdis.2015.01.002
- Iuzzini-Seigel, J., & Murray, E. (2017). Speech assessment in children with childhood apraxia of speech. *Perspectives of the ASHA Special Interest Groups*, 2(2), 47–60. https://doi.org/10. 1044/persp2.SIG2.47
- Kent, R. D., Eichhorn, J., Wilson, E. M., Suk, Y., Bolt, D. M., & Vorperian, H. K. (2021). Auditory–perceptual features of speech in children and adults with Down syndrome: A speech profile analysis. *Journal of Speech, Language, and Hearing Research*, 64(4), 1157–1175. https://doi.org/10.1044/2021_JSLHR-20-00617
- Kent, R. D., & Vorperian, H. K. (2013). Speech impairment in Down syndrome: A review. *Journal of Speech, Language, and Hearing Research, 56*(1), 178–210. https://doi.org/10.1044/1092-4388(2012/12-0148)
- Lee, J., Hustad, K. C., & Weismer, G. (2014). Predicting speech intelligibility with a multiple speech subsystems approach in children with cerebral palsy. *Journal of Speech, Language, and Hearing Research*, 57(5), 1666–1678. https://doi.org/10.1044/ 2014_JSLHR-S-13-0292
- Levy, E. S., Chang, Y. M., Hwang, K., & McAuliffe, M. J. (2021). Perceptual and acoustic effects of dual-focus speech treatment in children with dysarthria. *Journal of Speech, Language, and Hearing Research, 64*(6S), 2301–2316. https://doi. org/10.1044/2020_JSLHR-20-00301
- Lewis, B. A., Freebairn, L. A., Hansen, A. J., Iyengar, S. K., & Taylor, H. G. (2004). School-age follow-up of children with

childhood apraxia of speech. *Language, Speech, and Hearing Services in Schools, 35*(2), 122–140. https://doi.org/10.1044/0161-1461(2004/014)

- Maas, E., Butalla, C. E., & Farinella, K. A. (2012). Feedback frequency in treatment for childhood apraxia of speech. *American Journal of Speech-Language Pathology*, 21(3), 239–257. https:// doi.org/10.1044/1058-0360(2012/11-0119)
- Maas, E., Gildersleeve-Neumann, C., Jakielski, K., Kovacs, N., Stoeckel, R., Vradelis, H., & Welsh, M. (2019). Bang for your buck: A single-case experimental design study of practice amount and distribution in treatment for childhood apraxia of speech. *Journal of Speech, Language, and Hearing Research*, 62(9), 3160–3182. https://doi.org/10.1044/2019_JSLHR-S-18-0212
- Marquardt, T. P., Jacks, A., & Davis, B. L. (2004). Token-totoken variability in developmental apraxia of speech: Three longitudinal case studies. *Clinical Linguistics & Phonetics*, 18(2), 127–144. https://doi.org/10.1080/02699200310001615050
- McCabe, P., Thomas, D. C., & Murray, E. (2020). Rapid syllable transition treatment—A treatment for childhood apraxia of speech and other pediatric motor speech disorders. *Perspectives* of the ASHA Special Interest Groups, 5(4), 821–830. https://doi. org/10.1044/2020_PERSP-19-00165
- McHenry, M. A. (1999). Aerodynamic, acoustic, and perceptual measures of nasality following traumatic brain injury. *Brain Injury*, 13(4), 281–290. https://doi.org/10.1080/026990599121656
- Mei, C., Reilly, S., Bickerton, M., Mensah, F., Turner, S., Kumaranayagam, D., Pennington, L., Reddihough, D., & Morgan, A. (2020). Speech in children with cerebral palsy. *Developmental Medicine and Child Neurology*, 62(12), 1374–1382. https://doi.org/10.1111/dmcn.14592
- Mei, C., Reilly, S., Reddihough, D., Mensah, F., & Morgan, A. (2014). Motor speech impairment, activity, and participation in children with cerebral palsy. *International Journal of Speech-Language Pathology*, 16(4), 427–435. https://doi.org/10.3109/ 17549507.2014.917439
- Miller, H. E., Ballard, K. J., Campbell, J., Smith, M., Plante, A. S., Aytur, S. A., & Robin, D. A. (2021). Improvements in speech of children with apraxia: The efficacy of Treatment for Establishing Motor Program Organization (TEMPOSM). *Developmental Neurorehabilitation*, 24(7), 494–509. https://doi. org/10.1080/17518423.2021.1916113
- Morgan, A. T., & Liégeois, F. (2010). Re-thinking diagnostic classification of the dysarthrias: A developmental perspective. *Folia Phoniatrica et Logopaedica*, 62(3), 120–126. https://doi. org/10.1159/000287210
- Murray, E., Iuzzini-Seigel, J., Maas, E., Terband, H., & Ballard, K. J. (2021). Differential diagnosis of childhood apraxia of speech compared to other speech sound disorders: A systematic review. *American Journal of Speech-Language Pathology*, 30(1), 279–300. https://doi.org/10.1044/2020_AJSLP-20-00063
- Murray, E., McCabe, P., Heard, R., & Ballard, K. J. (2015). Differential diagnosis of children with suspected childhood apraxia of speech. *Journal of Speech, Language, and Hearing Research,* 58(1), 43–60. https://doi.org/10.1044/2014_JSLHR-S-12-0358
- Nijland, L., Maassen, B., Meulen, S. V. D., Gabreëls, F., Kraaimaat, F. W., & Schreuder, R. (2002). Coarticulation patterns in children with developmental apraxia of speech. *Clini*cal Linguistics & Phonetics, 16(6), 461–483. https://doi.org/10. 1080/02699200210159103
- Nittrouer, S., Studdert-Kennedy, M., & McGowan, R. S. (1989). The emergence of phonetic segments: Evidence from the spectral structure of fricative-vowel syllables spoken by children and adults. *Journal of Speech and Hearing Research*, 32(1), 120–132. https://doi.org/10.1044/jshr.3201.120

- Noiray, A., Abakarova, D., Rubertus, E., Krüger, S., & Tiede, M. (2018). How do children organize their speech in the first years of life? Insight from ultrasound imaging. *Journal of Speech, Language, and Hearing Research, 61*(6), 1355–1368. https://doi.org/10.1044/2018_JSLHR-S-17-0148
- Overby, M. S., & Caspari, S. S. (2015). Volubility, consonant, and syllable characteristics in infants and toddlers later diagnosed with childhood apraxia of speech: A pilot study. *Journal of Communication Disorders*, 55, 44–62. https://doi.org/10. 1016/j.jcomdis.2015.04.001
- Overby, M. S., Caspari, S. S., & Schreiber, J. (2019). Volubility, consonant emergence, and syllabic structure in infants and toddlers later diagnosed with childhood apraxia of speech, speech sound disorder, and typical development: A retrospective video analysis. *Journal of Speech, Language, and Hearing Research*, 62(6), 1657–1675. https://doi.org/10.1044/2019_jslhr-s-18-0046
- Patel, R., Hustad, K. C., Connaghan, K. P., & Furr, W. (2012). Relationship between prosody and intelligibility in children with dysarthria. *Journal of Medical Speech-Language Pathol*ogy, 20(4), 17.
- Pennington, L., Miller, N., Robson, S., & Steen, N. (2010). Intensive speech and language therapy for older children with cerebral palsy: A systems approach. *Developmental Medicine & Child Neurology*, 52(4), 337–344. https://doi.org/10.1111/j. 1469-8749.2009.03366.x
- Pennington, L., Roelant, E., Thompson, V., Robson, S., Steen, N., & Miller, N. (2013). Intensive dysarthria therapy for younger children with cerebral palsy. *Developmental Medicine & Child Neurology*, 55(5), 464–471. https://doi.org/10.1111/dmcn.12098
- Preston, J. L., Benway, N. R., Leece, M. C., & Caballero, N. F. (2021). Concurrent validity between two sound sequencing tasks used to identify childhood apraxia of speech in schoolage children. *American Journal of Speech-Language Pathol*ogy, 30(3S), 1580–1588. https://doi.org/10.1044/2020_AJSLP-20-00108
- Preston, J. L., Leece, M. C., & Maas, E. (2016). Intensive treatment with ultrasound visual feedback for speech sound errors in childhood apraxia. *Frontiers in Human Neuroscience*, 10, 440. https://doi.org/10.3389/fnhum.2016.00440
- Preston, J. L., Leece, M. C., McNamara, K., & Maas, E. (2017). Variable practice to enhance speech learning in ultrasound biofeedback treatment for childhood apraxia of speech: A single case experimental study. *American Journal of Speech-Language Pathology*, 26(3), 840–852. https://doi.org/10.1044/ 2017_AJSLP-16-0155
- Randolph, J. (2005). Free-marginal multirater kappa: An alternative to Fleiss' fixed-marginal multirater kappa. Joensuu University Learning and Instruction Symposium, October 14–15, 2005, Joensuu, Finland.
- Rosenbek, J. C., & Wertz, R. T. (1972). A review of fifty cases of developmental apraxia of speech. *Language, Speech, and Hearing Services in Schools, 3*(1), 23–33. https://doi.org/10. 1044/0161-1461.0301.23
- Rvachew, S., Hodge, M., & Ohberg, A. (2005). Obtaining and interpreting maximum performance tasks from children: A tutorial. Journal of Speech-Language Pathology and Audiology, 29(4), 146.
- Schölderle, T., Haas, E., & Ziegler, W. (2020). Age norms for auditory–perceptual neurophonetic parameters: A prerequisite for the assessment of childhood dysarthria. *Journal of Speech, Language, and Hearing Research,* 63(4), 1071–1082. https:// doi.org/10.1044/2020_JSLHR-19-00114
- Shriberg, L. D., Aram, D. M., & Kwiatkowski, J. (1997). Developmental apraxia of speech: I. Descriptive and theoretical

perspectives. Journal of Speech, Language, and Hearing Research, 40(2), 273–285. https://doi.org/10.1044/jslhr.4002. 273

- Shriberg, L. D., Campbell, T. F., Karlsson, H. B., Brown, R. L., McSweeny, J. L., & Nadler, C. J. (2003). A diagnostic marker for childhood apraxia of speech: The lexical stress ratio. *Clinical Linguistics & Phonetics*, 17(7), 549–574. https://doi.org/10. 1080/0269920031000138123
- Shriberg, L. D., Lohmeier, H. L., Strand, E. A., & Jakielski, K. J. (2012). Encoding, memory, and transcoding deficits in childhood apraxia of speech. *Clinical Linguistics & Phonetics*, 26(5), 445–482. https://doi.org/10.3109/02699206.2012.655841
- Shriberg, L. D., Potter, N. L., & Strand, E. A. (2011). Prevalence and phenotype of childhood apraxia of speech in youth with galactosemia. *Journal of Speech, Language, and Hearing Research*, 54(2), 487–519. https://doi.org/10.1044/1092-4388(2010/10-0068)
- Shriberg, L. D., Strand, E. A., Fourakis, M., Jakielski, K. J., Hall, S. D., Karlsson, H. B., Mabie, H. L., McSweeny, J. L., Tilkens, C. M., & Wilson, D. L. (2017). A diagnostic marker to discriminate childhood apraxia of speech from speech delay: I. Development and description of the pause marker. *Journal of Speech, Language, and Hearing Research, 60*(4), S1096–S1117. https://doi.org/10.1044/2016_JSLHR-S-15-0296
- Shriberg, L. D., Strand, E. A., Jakielski, K. J., & Mabie, H. L. (2019). Estimates of the prevalence of speech and motor speech disorders in persons with complex neurodevelopmental disorders. *Clinical Linguistics & Phonetics*, 33(8), 707–736. https://doi.org/10.1080/02699206.2019.1595732
- Sigurdardottir, S., & Vik, T. (2011). Speech, expressive language, and verbal cognition of preschool children with cerebral palsy in Iceland. *Developmental Medicine and Child Neurology*, 53(1), 74–80. https://doi.org/10.1111/j.1469-8749.2010.03790.x
- Song, J. Y., Demuth, K., Shattuck-Hufnagel, S., & Ménard, L. (2013). The effects of coarticulation and morphological complexity on the production of English coda clusters: Acoustic and articulatory evidence from 2-year-olds and adults using ultrasound. *Journal of Phonetics*, 41(3–4), 281–295. https://doi. org/10.1016/j.wocn.2013.03.004
- Strand, E. A. (2020). Dynamic temporal and tactile cueing: A treatment strategy for childhood apraxia of speech. American Journal of Speech-Language Pathology, 29(1), 30–48. https:// doi.org/10.1044/2019 AJSLP-19-0005
- Strand, E. A., Duffy, J. R., Clark, H. M., & Josephs, K. (2014). The Apraxia of Speech Rating Scale: A tool for diagnosis and description of apraxia of speech. *Journal of Communication Disorders*, 51, 43–50. https://doi.org/10.1016/j.jcomdis.2014.06.008
- Strand, E. A., & McCauley, R. J. (2019). Dynamic Evaluation of Motor Speech Skill (DEMSS) manual. Brookes. https://doi. org/10.1016/j.jcomdis.2014.06.008
- Strand, E. A., McCauley, R. J., Weigand, S. D., Stoeckel, R. E., & Baas, B. S. (2013). A motor speech assessment for children with severe speech disorders: Reliability and validity evidence. *Journal of Speech, Language, and Hearing Research*, 56(2), 505–520. https://doi.org/10.1044/1092-4388(2012/12-0094)
- Terband, H., Namasivayam, A., Maas, E., van Brenk, F., Mailend, M. L., Diepeveen, S., van Lieshout, P., & Maassen, B. (2019). Assessment of childhood apraxia of speech: A review/tutorial of objective measurement techniques. *Journal of Speech, Language, and Hearing Research, 62*(8S), 2999–3032. https://doi.org/10.1044/2019_jslhr-s-csmc7-19-0214
- Thoonen, G., Maassen, B., Gabreels, F., & Schreuder, R. (1999). Validity of maximum performance tasks to diagnose motor speech disorders in children. *Clinical Linguistics & Phonetics*, 13(1), 1–23. https://doi.org/10.1080/026992099299211

- Thoonen, G., Maassen, B., Wit, J., Gabreels, F., & Schreuder, R. (1996). The integrated use of maximum performance tasks in differential diagnostic evaluations among children with motor speech disorders. *Clinical Linguistics & Phonetics*, 10(4), 311–336. https://doi.org/10.3109/02699209608985178
- Timmins, C., Hardcastle, W., McCann, J., Wood, S., & Wishart, J. (2008). Coarticulation in children with Down's syndrome: An electropalatographic analysis. 8th International Seminar on Speech Production, 273–276. http://issp2008.loria.fr/Proceedings/PDF/ issp2008-62.pdf
- Tjaden, K., & Wilding, G. E. (2005). Effect of rate reduction and increased loudness on acoustic measures of anticipatory coarticulation in multiple sclerosis and Parkinson's disease. *Journal* of Speech, Language, and Hearing Research, 48(2), 261–277. https://doi.org/10.1044/1092-4388(2005/018)
- van Doorn, J. V., & Sheard, C. (2001). Fundamental frequency patterns in cerebral palsied speech. *Clinical Linguistics & Phonetics*, 15(7), 585–601. https://doi.org/10.1080/02699200110078113
- Vuolo, J., & Goffman, L. (2018). Language skill mediates the relationship between language load and articulatory variability in children with language and speech sound disorders. *Journal* of Speech, Language, and Hearing Research, 61(12), 3010–3022. https://doi.org/10.1044/2018_JSLHR-L-18-0055
- Wilson, E. M., Abbeduto, L., Camarata, S. M., & Shriberg, L. D. (2019). Estimates of the prevalence of speech and motor

speech disorders in adolescents with Down syndrome. *Clinical Linguistics & Phonetics*, 33(8), 772–789. https://doi.org/10. 1080/02699206.2019.1595735

- Wong, E. C., Velleman, S. L., Tong, M. C., & Lee, K. Y. (2021). Pitch variation in children with childhood apraxia of speech: Preliminary findings. *American Journal of Speech-Language Pathology*, 30(3S), 1511–1524. https://doi.org/10.1044/2021_AJSLP-20-00150
- Workinger, M. S., & Kent, R. D. (1991). Perceptual analysis of the dysarthrias in children with athetoid and spastic cerebral palsy. In C. A. Moore, K. M. Yorkston, & D. R. Beukelman (Eds.), *Dysarthria and apraxia of speech: Perspectives on management* (pp. 109–126). Brookes.
- Zharkova, N., Hewlett, N., & Hardcastle, W. J. (2011). Coarticulation as an indicator of speech motor control development in children: An ultrasound study. *Motor Control*, 15(1), 118–140. https://doi.org/10.1123/mcj.15.1.118
- Zharkova, N., Hewlett, N., & Hardcastle, W. J. (2012). An ultrasound study of lingual coarticulation in /sV/ syllables produced by adults and typically developing children. *Journal of the International Phonetic Association*, 42(2), 193–208. https://doi.org/ 10.1017/S0025100312000060
- Ziegler, W. (2002). Task-related factors in oral motor control: Speech and oral diadochokinesis in dysarthria and apraxia of speech. *Brain and Language*, 80(3), 556–575. https://doi.org/ 10.1006/brln.2001.2614