JSLHR

Research Note

Poor Speech Perception Is Not a Core Deficit of Childhood Apraxia of Speech: Preliminary Findings

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Purpose: Childhood apraxia of speech (CAS) is hypothesized to arise from deficits in speech motor planning and programming, but the influence of abnormal speech perception in CAS on these processes is debated. This study examined speech perception abilities among children with CAS with and without language impairment compared to those with language impairment, speech delay, and typically developing peers.
Method: Speech perception was measured by discrimination of synthesized speech syllable continua that varied in frequency (/dɑ/–/gɑ/). Groups were classified by performance on speech and language assessments and compared on syllable discrimination thresholds. Within-group variability was also evaluated.
Results: Children with CAS without language impairment did not significantly differ in syllable discrimination compared to typically developing peers. In contrast, those with CAS and language impairment showed significantly poorer syllable discrimination abilities compared to children with CAS only and typically developing peers. Children with speech delay and language impairment also showed significantly poorer discrimination abilities, with appreciable within-group variability.
Conclusions: These findings suggest that speech perception deficits are not a core feature of CAS but rather occur with co-occurring language impairment in a subset of children with CAS. This study establishes the significance of accounting for language ability in children with CAS.
Supplemental Materials: https://doi.org/10.23641/asha.5848056

Childhood apraxia of speech (CAS) is a pediatric speech disorder that occurs in approximately one to two children per thousand (Shriberg, Aram, & Kwiatkowski, 1997). CAS often results in a severe communication impairment that can have lasting effects on social, academic, and vocational functioning (Bird, Bishop, & Freeman, 1995; Felsenfeld, Broen, & McGue, 1992; Rice, Sell, & Hadley, 1991; Silverman & Paulus, 1989). Speech abnormalities associated with CAS include inconsistent errors, disrupted transitions between sounds and syllables, and abnormal stress patterns (American Speech-Language-Hearing Association, 2007; Iuzzini & Forrest, 2010). Although the etiology of CAS has not been identified, these speech difficulties are nearly universally attributed to deficits in motor planning or programming (e.g., Iuzzini-Seigel, Hogan, Guarino, & Green, 2015; Nijland et al., 2003; Shriberg, Lohmeier, Strand, & Jakielski, 2012; Terband & Maassen, 2010). Planning and programming are central components of most theories of spoken word production (Guenther, Ghosh, & Tourville, 2006; Hickok, 2014; Levelt, 1989; Perkell, 1980; Van der Merwe, 2009) and are modeled as the processes that translate speech units (i.e., phones or syllables or words) into an ordered sequence of goals for the speech production mechanism. Presumably, during development, several factors could impair the establishment and maintenance of motor plans and programs. Proposed faulty mechanisms include poor speech sound perception, disrupted somatosensation of the tongue and palate, abnormally high levels of neural noise, atypical auditory–motor neural pathways, or overreliance on auditory feedback (Iuzzini-Seigel et al., 2015; Terband & Maassen, 2010). Additional research is needed to establish the validity of these putative mechanisms of CAS impairment; such knowledge will be essential for facilitating the ongoing effort to develop more targeted and efficacious treatments of this often persistent speech disorder.

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Editor: Julie Liss
Associate Editor: Maria Grigos
Received March 18, 2016
Revision received October 10, 2016
Accepted October 15, 2017
https://doi.org/10.1044/2017_JSLHR-S-16-0106

Disclosure: The authors have declared that no competing interests existed at the time of publication.

The current investigation tests one potential causal mechanism that could disrupt the development of normal speech motor planning and programming processes: impaired speech sound perception (Maassen, Groenen, & Crul, 2003; Nijland, 2009; Shriberg et al., 2012). Empirical studies have explicated the importance of early speech perception on later emerging speech production (Eimas, Siqueland, Jusczyk, & Vigorito, 1971; Jusczyk & Hohne, 1997; Kuhl, 1993; Tsao, Liu, & Kuhl, 2004), and speech perception difficulties have been reported in a small number of studies on children with CAS. Specifically, children with CAS have shown poor auditory discrimination relative to typically developing controls of the following: vowels (Maassen et al., 2003), consonants contrasting in place of articulation (Groenen, Maassen, Crul, & Thoonen, 1996; Hoit-Dalgaard, Murry, & Kopp, 1983), voice onset time (Hoit-Dalgaard et al., 1983), and nonword discrimination (Bridgeman & Snowling, 1988; Nijland, 2009). Moreover, electroencephalographic investigations have also reported atypical neural responses to the presentation of oddball speech sounds within phonetic (voiced vs. unvoiced) and allophonic contrasts in children with CAS (Froud & Khamis-Dakwar, 2012). In contrast to these studies, others have reported no measurable differences between children with CAS and typically developing peers in speech perception tasks in identifying syllables that differ in place of articulation within the initial consonant (Nijland, 2009).

One potentially significant source of these mixed findings is varying language abilities among children with CAS. Studies reporting speech perception difficulties had not accounted for language impairment within the CAS groups (Froud & Khamis-Dakwar, 2012; Groenen et al., 1996; Maassen et al., 2003), whereas studies that excluded children with co-occurring language impairments reported no measurable differences between children with CAS and their typically developing peers on speech perception tasks (Groenen et al., 1996; Nijland, 2009). Co-occurring language impairments, characterized by difficulties with language formulation and comprehension (Leonard, 2014), are highly prevalent in children with CAS (Aram & Nation, 1982; Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004), and multiple studies have implicated speech perception deficits in children with language impairment (Elliott, Hammer, & Scholl, 1989; Frumkin & Rapin, 1980; Rosen, 2003; Stark & Heinz, 1996; Sussman, 1993, 2001; Tallal, Stark, Kallman, & Mellits, 1980). To our knowledge, no study has examined speech perception abilities in children with CAS without language impairment relative to that of children with CAS who have language impairment. This group contrast will help determine whether speech perception deficits are a core or co-occurring feature of CAS. Based on our careful review of the extant literature, we hypothesize that speech perception deficits will be found only in children with CAS who have language impairment.

Several studies have also identified speech perception deficits in children with speech delay (Edwards, Fox, & Rogers, 2002; Kenney, Barac-Cikoja, Finnegan, Jeffries, & Ludlow, 2006; Nijland, 2009; Rvachew & Jamieson, 1989), calling into question whether speech perception deficits are specific to CAS or represent a shared characteristic among children with a variety of speech production deficits. Therefore, an additional limitation of the extant work on speech perception deficits in children with CAS (Bridgeman & Snowling, 1988; Froud & Khamis-Dakwar, 2012; Groenen et al., 1996; Maassen et al., 2003) is the absence of a comparison group of children with disordered speech. Children with speech delay, characterized by delayed production of age-appropriate speech sounds (Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997), constitute an ideal non-CAS speech disordered comparison group (Iuzzini-Seigel et al., 2015; Nijland, 2009), as the speech production deficits associated with speech delay, relative to CAS, are predicted to manifest from distinct underlying bases (Shriberg et al., 2010). Specifically, weaknesses in speech perception in children with speech delay could be reflective of poorly specified phonemic representations, which have been implicated within this population (Munson, Edwards, & Beckman, 2005; Sutherland & Gillon, 2005). Taking these mixed findings of speech perception abilities in CAS and, more generally, speech delay into consideration, direct comparison of speech perception abilities in these populations is essential for identifying the distinct causal mechanisms that are hypothesized to underlie these disorders. We predict that children with speech delay will have speech perception difficulties compared to children with CAS without language impairment, which would indicate that speech perception is not globally linked to speech production impairments but is instead associated with speech delay and not CAS.

The current study evaluated speech perception abilities within five well-characterized groups: children with CAS relative to those with CAS and co-occurring language impairment, those with language impairment only, those with speech delay, and their typically developing peers. Our goal was to determine if speech perception deficits are central to the symptom profile of children with CAS or a symptom of the co-occurring language deficit and/or the presence of a nonspecific speech sound deficit (i.e., speech delay). Speech perception was assessed using a standard speech discrimination task of synthesized syllable pairs (/dɑ/–/gɑ/) that differ in place of articulation along a continuum varying in spectral structure. This is a common approach that has been characterized in typical development (e.g., Hazan et al., 2000) and previously shown to be sensitive to speech- and language-based impairments (Cabbage, 2013; Kraus et al., 1996). It remains unclear whether poor speech perception is associated with co-occurring language impairment in children with CAS because previous speech perception studies in children with CAS have included children with various language abilities. The current study will add to past literature by examining one aspect of speech perception (discriminating place of articulation) in children with CAS, with a focus on the impact of language ability. We hypothesize that poor speech perception will be linked to poor language skills in children with CAS, such that children with CAS and intact language abilities will not show difficulties in speech perception whereas those with CAS with language impairment will evidence poor speech perception. In accordance with previous literature, speech perception deficits are also anticipated for the majority of children with language impairment (Leonard, McGregor, & Allen, 1992; Sussman, 1993, 2001; Tallal et al., 1980) and speech delay (Broen, Strange, Doyle, & Heller, 1983; Edwards et al., 2002; Hoffman, Stager, & Daniloff, 1983; Kenney et al., 2006). If our hypotheses are supported, these preliminary findings will add to a growing body of work linking poor speech perception with language impairment and phonologically based speech production disorders (i.e., speech delay), but not with the core causal mechanism underlying CAS.

Method

Participants

Forty-seven children with typical and disordered speech and language ranging in age between 4;7 and 17;7 (years; months) participated in this study and were grouped as follows: children with CAS (n = 13; all male), children with language impairment only (n = 7; four male, three female), children with speech delay (n = 12; four male, eight female), and typically developing children (n = 15; eight male, seven female). These participants are part of a larger study on the biological basis of CAS (Centanni, Green, Iuzzini-Seigel, Bartlett, & Hogan, 2015; Centanni, Sanmann, et al., 2015; Iuzzini-Seigel et al., 2015). Participants were recruited from multiple sources, including research participant pools, speech-language pathologist referrals, and flyers posted to local schools, clinics, and parent groups. All participants were native monolingual English speakers. Group assignment was based on performance on a battery of standardized and customized assessments, treatment history, and parent report as described below.

Participants passed a pure-tone hearing screening at 20 dB for octave frequencies between 500 and 8000 Hz (American Speech-Language-Hearing Association, 1997). Moreover, participants evidenced normal nonverbal cognition based on a standard score of at least 75 on the Reynolds Intellectual Assessment Scales (RIAS; Reynolds & Kamphaus, 2003). In addition, none of the children enrolled evidenced signs of dysarthria. Children were administered the Sounds-in-Words subtest of the Goldman-Fristoe Test of Articulation–Second Edition (GFTA-2; Goldman & Fristoe, 2000) to measure speech articulation skills. We used a hybrid approach to broad transcription of the GFTA-2 in which we noted distortions. Transcriptions were performed by speech-language pathology students who were rigorously trained on disordered speech. Interrater reliability was calculated using the following formula: Agreements/(Agreements + Disagreements) × 100. The mean level of agreement was 89%, a high level of agreement between raters for transcription of vowels and consonants for disordered speech. To measure language skills, the core subtests of the Clinical Evaluation of Language Fundamentals–Fourth Edition (CELF-4; Semel, Wiig, & Secord, 2003) were administered. For a subset of children with typical language abilities with no concerns about language development as noted by parent report, the Clinical Evaluation of Language Fundamentals–Fourth Edition Screening Test (CELF-4 Screening Test; Semel, Wiig, & Secord, 2004) was administered instead of the full CELF-4, because the CELF-4 Screening Test has high sensitivity and, in particular, specificity for the identification of language impairment versus typical language development (Semel et al., 2004). To prevent bias against participants with speech sound production errors during evaluation of language abilities, children received credit for responses in which they demonstrated an appropriate grammatical response even if they did not pronounce it correctly. For instance, if a child used a vocalic syllable to mark the present participle "-ing" or if a child used a substitution to mark a plural or possessive /s/, the child was given credit for these responses.

Group Assignment

Participants were assigned to the typically developing group (n = 15) if they met all the criteria listed above, scored above the 16th percentile on the GFTA-2, and exhibited normal language based on a standard Core Language score of 88 or higher on the CELF-4 or a score of "Pass" on the CELF-4 Screening Test. In addition, typically developing children had no history of speech and language treatment to prevent inclusion of participants with remediated speech and/or language deficits.

Group assignment to the speech sound disordered groups was determined by a protocol described in previous research (Iuzzini-Seigel et al., 2015). Children were assigned to the speech sound disordered groups if they scored at or below the 16th percentile on the GFTA-2. They were further differentially diagnosed based on diagnostic and treatment history, rating of CAS features, and language ability. Of the children with low performance on the GFTA-2, eight children were referred with a diagnosis of CAS by a speech-language pathologist with expertise in CAS, and one was reported to have CAS per parent report.

Although children were referred with a CAS diagnosis, assignment to the CAS group in this study required that participants evidenced at least five out of 11 CAS features (adapted from Shriberg, Potter, & Strand, 2011) while completing the GFTA-2. Shriberg et al. (2011) have previously assessed these features across three contexts that vary in difficulty. Because previous work (Iuzzini-Seigel, Hogan, & Green, 2017; Nijland, 2009) shows that children with CAS, typical development, and speech delay evidence increased errors and speech inconsistency on single words, this study conducted analysis of clearly defined CAS features on words from the GFTA-2. If a child evidenced a feature one or more times, it was counted as being positive for that feature, and any combination of five or more features during production of this simple assessment yielded assignment to the CAS group. This procedure was designed to prevent overdiagnosis, an ongoing clinical issue (e.g., Murray, McCabe, Heard, & Ballard, 2015) that is reportedly more prevalent than underdiagnosis (e.g., Davis, Jakielski, & Marquardt, 1998; Murray et al., 2015). The CAS characteristics assessed included vowel distortions, difficulty in achieving initial articulatory or coarticulatory configurations, equal stress or lexical stress errors, distorted substitutions, syllable segregation, groping, intrusive schwa, voicing errors, slow rate, increased difficulty with longer words (e.g., bath compared with bathtub), and disturbed resonance. Operational definitions were created for these characteristics, and two speech language pathologists with expertise in CAS blind-rated each child's GFTA-2 productions (see Appendix for operational definitions; Iuzzini-Seigel et al., 2017). Interrater reliability for CAS feature ratings was calculated on GFTA-2 responses from all participants in the sample. The intraclass correlation coefficient with absolute error in parenthesis was .93 (± .6), a high level of agreement for perceptual feature rating using the operational definitions. An overview of individual features evidenced by each participant is provided in Supplemental Material S1. Using these criteria, a CAS diagnosis was confirmed in 13 participants, who evidenced six features on average.

Children with a confirmed diagnosis of CAS ($n = 13$) were then categorized into the CAS subgroups with and without language impairment. Children were assigned to the CAS group without language impairment ($n = 7$) if they evidenced five or more features and exhibited normal language based on a standard Core Language score of 88 or higher on the CELF-4. Children were assigned to the CAS with language impairment group ($n = 6$) if they evidenced five or more CAS features and low language performance on the CELF-4 (< 88).

Children were assigned to the speech delay group ($n = 12$) if they produced fewer than five CAS characteristics and evidenced normal language. In addition, children were excluded from the speech delay group if, per parent report, they had a history of treatment for CAS; this criterion was used to prevent the erroneous assignment of children with partially resolved CAS to the speech delay group. Children in the speech delay group evidenced an average of two CAS features (see Table 1) and demonstrated consistent production errors primarily with one to two of the "Late 8" speech sounds (Shriberg, 1993), which largely consisted of /s/, /z/, and/or /r/.

Children were assigned to the group with language impairment ($n = 7$) if they evidenced low language performance on the CELF-4 (< 88) and if they scored above the 16th percentile on the GFTA-2 and evidenced fewer than five CAS features. On average, children in the group with language impairment evidenced one CAS feature.

Stimuli

A 41-step synthesized speech continuum was developed for the syllable discrimination task using a Klatt synthesizer (Klatt, 1980). Syllable stimuli along the continuum only differed in F3 onset frequency, which ranged from 1920 Hz (/ɡɑ/) to 2700 Hz (/dɑ/). The onset frequency increased by 20 Hz for each step in the continuum. For all stimuli, syllable duration was set at 250 ms, including the 45 ms formant transition period into the steady state of the vowel /ɑ/.

Procedure

Stimuli were presented on a desktop computer through a custom-designed software program utilizing parameter estimation by sequential tracking (PEST) to dynamically determine the just noticeable difference between stimuli for each participant, as designed and implemented in previous published studies (Carrell, Bradlow, Nicol, Koch, & Kraus, 1999; Kraus et al., 1996; Taylor & Creelman, 1967). Children were either fitted with insert earphones (Etymotic ER-1A) or noise-canceling headphones (Sennheiser HD280Pro) for task presentation.

Stimuli were presented in a dual-pair, or 4IAX, discrimination task within the PEST paradigm (Carrell et al., 1999; Kraus et al., 1996). For each trial, two discrete pairs of stimuli were presented: a reference pair and an experimental pair. The reference pair contained two identical syllables from the /dɑ/ end of the continuum. The experimental pair contained two different stimuli including the /dɑ/ syllable from the reference pair and a second syllable—from along the continuum—which was dynamically determined by the PEST algorithm. The experimental pairs started with stimuli that were highly contrastive and then distinctiveness between stimuli systematically decreased or increased based on the accuracy of a participant's response. The task continued in this manner until a threshold of discrimination along the continuum was achieved, in which the participant reliably reached an overall accuracy of 69% correct (as has been defined by signal detection theory for this type of discrimination task; Green & Swets, 1966).

Initially, a short training phase with different stimuli confirmed understanding of the task and ability to perform the task without exposure to the /dɑ/ and /ɡɑ/ stimuli. At the start of each trial, the computer screen displayed two unlit light bulbs, labeled "1" and "2." At the auditory presentation of each stimulus pair, the corresponding light bulb flashed concurrently. Children were asked to determine which pair contained the different stimuli and indicate their choice via a two-button handheld response box. The PEST algorithm controlled presentation such that the experimental pair was randomized and counterbalanced over the two buttons across all trials for all participants. The light bulb corresponding to the child's selection illuminated synchronously with the button press. After 750 ms, children were provided feedback via the illumination of the light bulb corresponding to the correct answer as reinforcement to keep children actively engaged throughout the task, per the PEST paradigm protocol as previously described (Carrell et al., 1999). After a response or after 3,500 ms if the child had not responded, both light bulbs flashed once to signal the start of the next trial. With this dynamic method of assessment, the number of trials required to determine the threshold of discrimination varied across participants, ranging from six to 66 trials.

Table 1. Participant demographics by group.

Demographic factors	Typically developing (*n* = 15)	CAS with no language impairment (*n* = 7)	CAS with language impairment (*n* = 6)	Language impairment (*n* = 7)	Speech delay (*n* = 12)	*F* (max *df* = 4.42)
Age (in months)	117.31 ± 31.01 (94–212)	121.57 ± 50.29 (55–207)	114.16 ± 31.24 (64–148)	122.14 ± 16.38 (92–144)	101.42 ± 12.64 (76–119)	0.51
RIAS SS	118.13 ± 15.56 (89–142)	110.71 ± 5.85 (99–115)	106.00 ± 16.02 (81–123)	101.71 ± 3.45 (97–107)	113.58 ± 16.27 (84–141)	2.09
GFTA-2 Percentile	42.13 ± 12.89 (21st–65th)	5.00 ± 5.38 (1st–15th)	2.67 ± 1.75 (1st–6th)	36.00 ± 10.78 (19th–56th)	4.42 ± 6.26 (1st–16th)	32.95*
CELF-4 Core SS	112.83 ± 15.04 (93–136)	106.29 ± 14.53 (90–133)	68.67 ± 17.36 (44–85)	74.71 ± 10.94 (58–87)	105.63 ± 16.02 (88–129)	12.11*
No. of CAS features^{+}	0.90 ± 0.70 (0–2)	6.00 ± 1.22 (5–8)	5.40 ± 1.14 (5–7)	1.30 ± 0.81 (0–2)	2.40 ± 0.99 (0–4)	—

Note. Mean ± standard deviation displayed with range in parentheses, by group. CAS = childhood apraxia of speech; GFTA-2 = Goldman-Fristoe Test of Articulation–Second Edition (Goldman & Fristoe, 2000); CELF-4 = Clinical Evaluation of Language Fundamentals–Fourth Edition (Semel et al., 2004; note that 14 children received a score of "Pass" on the Clinical Evaluation of Language Fundamentals–Fourth Edition Screening Test and therefore did not complete the full CELF-4 assessment, so these children are not included within the CELF scores or corresponding statistics reported in this table); RIAS = Reynolds Intellectual Assessment Scales (Reynolds & Kamphaus, 2003); SS = standard score; $^{+}$ = feature list and assessment procedure adapted from Shriberg et al. (2011).

*$p < .001$.

Statistical Analyses

One-way analyses of variance were used to test for group differences in age, nonverbal IQ, articulation (as indicated by the GFTA-2), or core language abilities (as indicated by the CELF-4). Post hoc comparisons were performed using false discovery rate (FDR) adjustments (i.e., controlling for the FDR; Benjamini, Drai, Elmer, Kafkafi, & Golani, 2001).

Syllable discrimination thresholds were examined by group to verify whether the assumption of normality was met using the Shapiro–Wilk test. The assumption of normality was not met in all groups; therefore, group effects in syllable discrimination were tested using nonparametric (Kruskal–Wallis) approaches. Post hoc comparisons were then performed using the Dunn–Bonferroni adjusted significance levels for multiple comparisons (Dunn, 1964). To check for outliers, each participant's syllable discrimination score was expressed as a within-group *z* score. Based on this transform, four participants were identified as outliers because their discrimination thresholds were 3 *SD*s above or below the mean discrimination threshold (CAS without language impairment: $n = 1$, CAS with language impairment: $n = 1$, typically developing: $n = 2$; Balota et al., 2007). Statistical analyses were conducted with and without these outliers.

To assess within-group variability, box plots were created with individual data points depicted, and the following measures of spread were characterized: interquartile range and 95% confidence intervals.

Results

Participant Demographics

Table 1 provides data to support our well-characterized, carefully phenotyped participant groups: children with CAS, CAS with language impairment, language impairment, speech delay, and typically developing peers. One-way analyses of variance revealed no differences between groups in age, $F(4, 42) = 0.833$, $p = .512$, or nonverbal IQ, $F(4, 42) = 2.086$, $p = .10$. As expected, groups significantly differed in articulation as measured by the GFTA-2, $F(4, 42) = 32.954$, $p < .001$, and core language abilities as indicated by the CELF-4, $F(4, 29) = 12.107$, $p < .001$ (see Table 1 for an overview; note that children who passed the CELF-4 Screening Test as a CELF-4 equivalent were not included in the overview or analysis of language performance). Individual speech and language scores are provided in Supplemental Material S2. FDR-adjusted post hoc tests showed that the groups with CAS without language impairment, CAS with language impairment, and speech delay groups did not significantly differ in articulation accuracy (as indicated by the GFTA-2) and performed more poorly than those with language impairment and typical development ($p < .001$), who did not differ in articulation performance. For the CELF-4 core language measure, post hoc tests demonstrated that children with CAS without language impairment, speech delay, and typical development did not significantly differ and in turn performed significantly better than those with CAS with language impairment and those with language impairment ($p < .005$), who did not significantly differ in language performance.

Syllable Discrimination Task

Perception was measured through a syllable discrimination task composed of synthesized syllable pairs characterized by spectral changes that distinguish place of articulation between /dɑ/–/gɑ/. Speech production accuracy was verified by the research team to ensure that all participants evidenced reliable production of the phonemes /d/ and /g/ (as indicated by the GFTA-2), thereby ruling out production difficulty as a possible confounding factor in measuring discrimination of these sounds.

The nonparametric statistical analysis revealed several group differences in discrimination of the /dɑ/–/gɑ/ contrast (see Figure 1). With the outliers included, the Kruskal–Wallis test resulted in significant differences between groups ($n = 47$, $p = .002$). Post hoc comparisons showed that the group with CAS with no language impairment did not significantly differ from the typically developing group, and both of these groups showed significantly better discrimination thresholds than children with CAS with language impairment ($p < .05$). Children in both the language impairment and speech delay groups did not significantly differ from any other groups. Following removal of four outliers, significant group differences in discrimination of the /dɑ/–/gɑ/ contrast remained (Kruskal–Wallis test; $n = 43$, $p = .002$). Post hoc group comparisons showed that the group with CAS with no language impairment did not significantly differ from the typically developing group, and both of these groups showed significantly better discrimination thresholds than children with speech delay ($p < .05$) and CAS with language impairment ($p < .005$). Discrimination thresholds of children with language impairment did not significantly differ from the typically developing group or groups with CAS (with and without language impairment).

Figure 1. Syllable discrimination thresholds by group. Within-group box plots of syllable discrimination thresholds for the /dɑ/–/gɑ/ continuum. Each box plot illustrates median, interquartile range (in gray), 95% confidence intervals around the median, and individual values (circles). Higher discrimination thresholds indicate poorer performance. The significance map (top left corner) displays the statistically significant ($p < .05$) post hoc comparisons between groups with outliers excluded. CAS = childhood apraxia of speech.

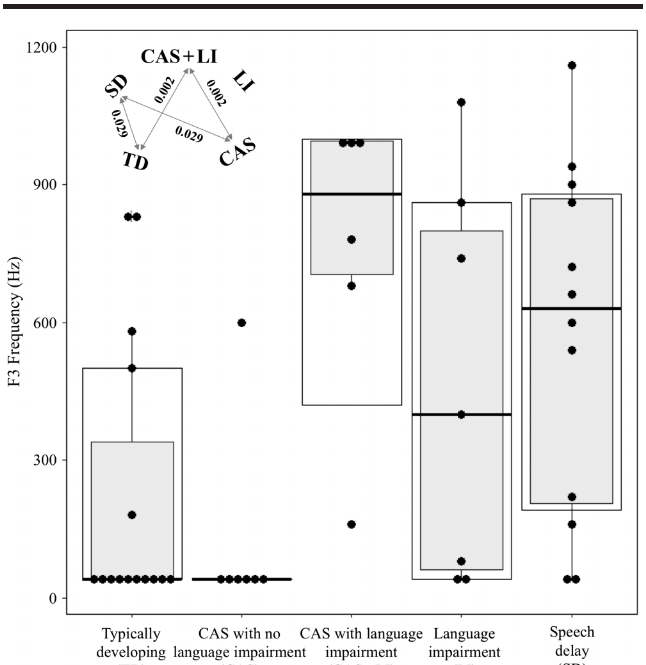

Figure 1 highlights the clear differences that were observed in within-group variability in syllable discrimination. Based on the interquartile ranges and 95% confidence intervals depicted by group, children with CAS with no language impairment were highly homogenous, whereas those with CAS with language impairment were moderately variable. By comparison, children with language impairment and speech delay demonstrated substantial within-group variability, for the spread within each of these groups was approximately twice as large as that of the typically developing group.

Although these groups did not significantly differ in age, a broad age range is evident within the present sample. Therefore, Pearson correlation analyses confirmed that there are no significant connections between age and syllable discrimination threshold in the sample as a whole ($r = -.253$, $p = .102$) or within any group (on average, $r = -.344$, range: -0.548 to -0.176, $p > .2$).

Discussion

In this study, we compared the speech perception abilities of children with CAS with and without language impairment compared to children with language impairment, speech delay, and typical development. As predicted, children with CAS and normal language showed age-appropriate syllable discrimination skills, whereas most of the children with co-occurring language impairment had poor discrimination skills. By comparison, children with speech delay and those with language impairment showed appreciable within-group variability. Overall, these findings suggest that speech perception deficits in children with CAS are associated with co-occurring language impairment and are not a core deficit of CAS.

Poor Speech Perception Is Associated With Language Impairment in Children With CAS

Although the literature to date reports a wide range of perceptual deficits in children with CAS (Groenen et al., 1996; Hoit-Dalgaard et al., 1983), our data suggest that previous mixed findings may be due to the lack of experimental control for co-occurring language deficits. Our observation of normal perceptual skills is, however, consistent with the one available study that excluded children with co-occurring language impairment from their sample (Nijland, 2009). These findings underscore the importance of accounting for language abilities in research on CAS, because it is possible that language abilities contribute to the varying symptom profiles and treatment outcomes that have been commonly observed among children with CAS.

The present findings of age-appropriate speech perception skills in children with CAS with no language impairment have implications for identifying the core deficits that underlie CAS. First, these findings do not support the notion that impaired speech perception is a core deficit in CAS. Alternatively, the relatively strong auditory perceptual abilities of these children with CAS align with previous findings that children with CAS rely heavily on auditory feedback during speech production (Iuzzini-Seigel et al., 2015). Thus, the present findings suggest that poor speech perception is not a factor contributing to the difficulties in motor planning or programming associated with CAS.

Variable Speech Perception in Children With Language Impairments

In the current study, many, but not all, of the children in the language-impaired group had weak speech perception skills, which is consistent with prior research showing high degrees of variation among children with language impairment in speech perception skills, such as phonetic and syllabic discrimination (Elliott et al., 1989; Frumkin & Rapin, 1980; Stark & Heinz, 1996; Sussman, 1993, 2001; Tallal et al., 1980). Yet, it remains unclear whether there may be specific profiles of language impairment that may explain this variability in speech perception. The majority of studies in this area investigated children with mixed receptive and expressive impairments (e.g., Evans, Viele, Kass, & Tang, 2002; Stark & Heinz, 1996; Sussman, 1993; Tallal et al., 1980), though some focused on receptive impairment and did not characterize expressive language abilities (e.g., Elliott et al., 1989; Sussman, 2001) and others did not distinguish between receptive and expressive language (e.g., Frumkin & Rapin, 1980; Rosen, 2003).

In contrast, all of the children with CAS with language impairment exhibited significantly poorer speech discrimination skills than did both typically developing children and those with CAS without language impairment. These findings suggest that language impairment is associated with considerable within-group variability, and co-occurring language impairment in individuals with CAS may be a significant contributing factor to the heterogeneous symptom profile in children with CAS.

Variable Speech Perception in Children With Speech Delay

A link between deficient speech perception and delayed speech production (Broen et al., 1983; Edwards et al., 2002; Hoffman, Daniloff, Bengoa, & Schuckers, 1985; Kenney et al., 2006; Nijland, 2009; Rvachew & Jamieson, 1989) has been proposed in prior studies, which supports the notion that deficient speech perception leads to weak phonologic representations and, ultimately, speech errors (Hoffman et al., 1985; Nijland, 2009; Rvachew & Jamieson, 1989). However, speech perception deficits have not been consistently identified in children with speech delays (Lof & Synan, 1997). Inclusion of a non-CAS speech delay group in this study allowed for direct comparison of speech perception abilities in children with CAS relative to other speech-specific production deficits, thereby addressing a limitation of previous research (Bridgeman & Snowling, 1988; Froud & Khamis-Dakwar, 2012; Groenen et al., 1996; Maassen et al., 2003). Group-level analyses indicated that children with speech delay showed significantly poorer speech perception abilities than children with CAS with no language impairment. Yet, inspection of the spread across children with speech delay revealed substantial variability, similar to that of the group with language impairment; some children with speech delay showed speech perception difficulties where others did not. Because children with speech delay represent a range of speech errors and severity, further research is needed to determine what factors are associated with poor speech perception in this heterogeneous population. There continues to be a critical need to assess speech perception in children with speech delay and in those with language impairment and to consider the impact of individual variability in speech perception abilities on treatment outcomes.

Limitations and Future Directions

This study measured speech perception using only one acoustic parameter of speech perception. Future studies are needed to determine whether speech perception abilities in these groups may vary for other significant acoustic features, such as previously reported difficulties in perceiving vowels (Maassen et al., 2003) and voiceless consonants (Groenen et al., 1996; Hoit-Dalgaard et al., 1983) in children with CAS. In addition, these findings need to be replicated with a larger sample size while maintaining our conservative inclusion criteria to obtain well-selected, carefully phenotyped participant groupings.

Conclusions

Children with CAS with no language impairment demonstrated age-appropriate speech perception skills, whereas many, but not all, children with speech delay and language impairment evidenced poor speech perception. These preliminary findings suggest that speech perception deficits may not be a causal mechanism underlying CAS, but rather that speech perception seems to be associated with co-occurring language impairment that is present in a significant subset of children with CAS. Co-occurring language impairment may contribute to variable symptom profiles and treatment outcomes in children with CAS. This study establishes the importance of accounting for language ability among children with CAS when diagnosing and treating this population.

Acknowledgments

This research was supported by the University of Nebraska Health Research Consortium (Hogan & Green), the National Institutes of Health (R03 DC9667 to Hogan), the Childhood Apraxia of Speech Association of North America (Iuzzini), the Barkley Memorial Trust, and the National Institute of Health Institutional National Research Service Award (NIH T32 DC000038-22 to Zuk).

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Appendix

Operational Definitions for Childhood Apraxia of Speech Characteristics

1. **Vowel error:** A vowel production error in which the vowel is substituted for another phoneme OR in which the vowel is recognizable as a specific phoneme but it is not produced exactly correctly (e.g., not a prototypical production, may sound like it's in between two vowels). It is not considered an error if the vowel is substituted with another phoneme that is consistent with an adultlike model (e.g., /hɑt dag/ /hɑt dɔg/)
2. **Consonant distortion:** A consonant production error in which a speech sound is recognizable as a specific phoneme but it is not produced exactly correctly (e.g., an /s/ that is produced with lateralization or dentalization).
3. **Stress errors:** An error in which the appropriate stress is not produced correctly. For example: conDUCT versus CONduct have different stress patterns. It is considered an error if the stress is inappropriately equalized across syllables or on the wrong syllable.
4. **Syllable segregation:** Brief or lengthy pause between syllables, which is not appropriate.
5. **Groping:** Prevocalic (silent) articulatory searching prior to onset of phonation, possibly in an effort to improve the accuracy of the production. Video is needed to assess this feature.
6. **Intrusive schwa (e.g., in clusters):** A schwa is added in between consonants. For example, it may be inserted in between the consonants in a cluster (e.g., /blu/ becomes /bə̲lu/). This is NOT considered a "vowel error."
7. **Voicing errors:** 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 in between voicing categories (e.g., blurring of voicing boundaries).
8. **Slow rate:** Speech rate is not typical. It is slower during production of part (e.g., zzziiiiiiper/zipper) or the whole word (e.g., tooommmmaaatoooo/tomato).
9. **Increased difficulty with longer words:** The participant has a disproportionately increased number of errors as the number of syllables increases (as compared to words with fewer syllables).
10. **Resonance or nasality disturbance:** Sounds either *hyponasal*: not enough airflow out of nose/"stuffy" OR *hypernasal*: too much airflow out of nose for nonnasal phonemes (e.g., plosives).
11. **Difficulty in achieving initial articulatory configurations or transitionary movement gestures:** 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.