Bridging the Gap Between Speech and Language: Using Multimodal Treatment in a Child With Apraxia

Cheryl D. Tierney, MD, MPH,[a] Kathleen Pitterle, DO,[b] Marie Kurtz, MS, CCC-SLP,[c] Mark Nakhla,[d] Carlyn Todorow, MD[d]

Childhood apraxia of speech is a neurologic speech sound disorder in which children have difficulty constructing words and sounds due to poor motor planning and coordination of the articulators required for speech sound production. We report the case of a 3-year-old boy strongly suspected to have childhood apraxia of speech at 18 months of age who used multimodal communication to facilitate language development throughout his work with a speech language pathologist. In 18 months of an intensive structured program, he exhibited atypical rapid improvement, progressing from having no intelligible speech to achieving age-appropriate articulation. We suspect that early introduction of sign language by family proved to be a highly effective form of language development, that when coupled with intensive oro-motor and speech sound therapy, resulted in rapid resolution of symptoms.

Children with childhood apraxia of speech (CAS) have difficulty with coordination of the articulatory muscles required for speech, leading to decreased production of words and fewer vocalizations. CAS is further defined as a neurologic pediatric speech sound disorder in which precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits. The core impairment is in planning and/or programming spatiotemporal parameters of movement sequences, resulting in errors in speech sound production and prosody. The consensus-based core CAS features are "(a) inconsistent errors in consonants and vowels in repeated productions of syllables or words, (b) lengthened and disrupted co-articulatory transitions between sounds and syllables, and (c) inappropriate prosody, especially in the realization of lexical or phrasal stress."[1]

In children with speech delay and CAS, it is critical to intervene early to reduce long-term deficits in communication, social-emotional interactions, and later language learning.[2] The treatment time course for CAS can take years of therapy compared with other speech sound disorders due to underlying motor speech impairments. Slower progress also may be seen in cases with comorbidities, such as autism, fine motor delays, underlying language disorders and cognitive weaknesses, that may interfere with therapy or behavioral difficulties or emotional reactions to the communication difficulty.

Long-term data have been limited to support a specific treatment approach and how the method and intensity of treatment affects outcome.[3] Although there has been an increasing focus on treatment evidence for children with CAS, including some preliminary randomized control

abstract

[a]*Division of Pediatric Rehabilitation and Development, and Departments of* [b]*Internal Medicine-Pediatrics and* [c]*Orthopedics and Rehabilitation, Supervisor of Speech Language Pathology, Penn State Milton S. Hershey Medical Center, Hershey, Pennsylvania; and* [d]*The Penn State University College of Medicine, Hershey, Pennsylvania*

Dr Tierney conceptualized and designed this study, recruited the patient for this case report, facilitated the necessary observed meetings at the patient's speech language pathologist appointments, helped to draft the initial manuscript and all iterations thereafter and edited the final manuscript; Dr Pitterle helped to conceptualize and design this study, conducted a literature review for this case report, drafted the initial manuscript, and reviewed and revised the manuscript; Ms Kurtz, the speech language pathologist who worked with the patient described in this case report, met with both the patient and his mother several times during the drafting of this manuscript, contributed to the initial manuscript, and edited the final manuscript; Mr Nakhla helped to conceptualize and design this study, gathered clinical and medical data from the patient, the patient's mother, and the patient's speech language pathologist, reviewed the patient's chart and gathered appropriate medical history, helped to draft the initial manuscript, and edited the final manuscript; Ms Todorow gathered clinical and medical data from the patient's speech language pathologist, reviewed the patient's chart and gathered appropriate medical history, and edited the final manuscript; and all authors approved the final manuscript as submitted.

DOI: 10.1542/peds.2016-0007

Accepted for publication Jun 6, 2016

Address correspondence to Cheryl D. Tierney, MD, MPH, Division of Pediatric Rehabilitation and

To cite: Tierney CD, Pitterle K, Kurtz M, et al. Bridging the Gap Between Speech and Language: Using Multimodal Treatment in a Child With Apraxia. *Pediatrics.* 2016;138(3):e20160007

trials and nonrandomized control trials,[3,4] there remains a paucity of evidence for younger children with CAS. Numerous treatment interventions have been reported on an individual basis, including motor treatments, linguistic approaches, and augmentative and alternative communication (AAC) or combinations of these methods.[4–13] AAC allows children to build receptive language while giving them access to expressive language so that when speech develops these skills will carry over.[14] Most of the reviewed studies using AAC discussed outcomes with voice output devices or communication boards.[2,5,7,8,12] None of the 42 studies reviewed by Murray et al[5] used sign language as the primary AAC for children with CAS, yet the review of these studies suggested that AAC improved expressive language, reduced frustration, and resulted in transition to reliance on speech.

Parental concern exists that if a child is taught sign language it will dampen the development of speech, and the child will rely on signing rather than focusing on the more difficult task of motor speech development. For children with CAS, sign can be used successfully to augment and assist their communication as a bridge to speech development.[15] Children with CAS often develop their own gestural communication system, unless limited by fine and gross motor delays, and as a result sign language can be a natural and portable method of AAC.[14] Previous qualitative reports on the use of sign language, from parents and speech therapists, have shown an increase in speech attempts and reduced frustration expanding their expressive language. Sign language also provided encouragement and reinforcement for successful communication.[15] As oral skills and speech improved, sign language faded spontaneously.[16] More recent case reports, within the past 10 years on the use of sign language serving as AAC in CAS, were not found after an extensive literature review. Although older case reports discussed sign language as an AAC, our case report focused on the role of sign language in CAS as an effective bridge to natural speech.

CASE DESCRIPTION

Our patient is a 3-year-old boy who first presented with an expressive language delay at 8 months old with absent cooing and babbling. In addition, he showed difficulty with oro-motor control while eating, including near choking at 1 year of age due to poor mastication and excessive drooling. Due to expressive language delay, he underwent an audiology evaluation at 14 months, which was normal. He first received a modified barium swallow study at the age of 21 months in which swallow function was deemed normal but concerns for reflux, speech apraxia, and decreased chew function/tongue lateralization were all noted in the report. No other neurologic or developmental needs were identified, as noted in Table 1.

Due to maternal concern for language delay, his mother self-directed the initiation of sign language at 14 months. Signed Exact English was used to build vocabulary and expand expression to the multiword level. She noted that he readily gravitated toward using sign. At the time of his initial speech language pathologist appointment, he already used 50 to 60 signs to communicate, but natural speech had not progressed. Due to limited verbal language, he had little interest in playing with typical peers. Other AAC options were presented, such as a picture exchange communication system and a voice output device, but the patient continued to show a strong preference for sign.

At initial outpatient speech assessment, an oral motor evaluation was completed. He demonstrated mild drooling without evidence of any weakness or dysarthria. Drooling was assessed to be related to poor oral sensory awareness of saliva and decreased oral motor coordination to consistently manage secretions while engaged in speech, eating, or other attention-based tasks. He was noted to have poor labial protrusion in imitation but ability to functionally protrude lips in whistle and straw tasks. He was observed with open mouth posture at rest but ability to close on command. This skill increased in ease with tactile cuing (touch prompts). He demonstrated mildly asymmetric lip retraction with groping motion on command or when paired with voicing but adequate and typical retraction when

TABLE 1 Neurologic and Developmental History

Problem Identified	Assessment/Evaluation Completed	Results
Feeding difficulties	Upper gastrointestinal series (10/4/13)	Normal anatomy
	Rehabilitation swallow (5/9/13)	No aspiration, oro-motor deficits present
Expressive language delay	Multiple tympanograms (most recently 10/13/15)	Normal hearing sensitivity bilaterally
	Auditory brainstem response evaluation (3/8/13)	Normal hearing sensitivity from 500 to 4000 Hz
	Neurologic evaluation (8/5/13)	Normal neurologic examination
		No MRI performed
Recurrent otitis media	Bilateral myringotomy tubes placed (10/26/12)	
Fine motor delay		Identified and treated by occupational therapy
		Note: no gross motor delays identified

observed to smile in non–demand-based opportunities (spontaneous speech). These challenges when taken together support nonspeech oral apraxia. In addition, our patient demonstrated difficulties with voice initiation.

During single phoneme production tasks, he was observed with limited oral muscular differentiation producing minimal change in vowel productions. Stimulable vowel sounds were "uh," "ah," and "oh," but all required visual and tactile cuing and oral muscular groping was observed. In addition, stimulable consonants were limited to /b, d, m, g/, all of which were voiced. He was able to produce an approximation of uh-oh and inflection was described as "minimal."

Syllable/word imitation tasks were attempted. He was interested and attentive with good eye contact and noted attempts at many word productions. Speech attempts were characterized by sound separations, poor consonant–vowel blending, and overuse of the schwa vowel (phonetic sound "uh" in a word, such as the "uh" sound in the word "the"). Examples of early imitated words included "buh-o" for ball, "da" for dad, "muh" for mom, "guh" for go, and "m-o" for more.

Despite the noted challenges, he was able to use nonverbal gestures and facial expression to help communicate with others. His language scores as reported on the Pre-School Language Scale–5 were in the low-average range for expression and above-average range for comprehension. This testing profile indicates normal receptive language and likely normal cognition.

At 18 months of age, the diagnosis of CAS was highly suspected due to evidence consistent with the American Speech-Language-Hearing Association guidelines.[1] Our patient demonstrated inconsistent errors in consonant and vowel productions, poor co-articulatory transition from sounds to syllables, and deficits in prosody. Also present was evidence of motor groping and strong desire and effort to communicate without successful sound production. Although the rapidity of our patient's speech improvement before the age of 3 may have suggested constitutional speech delay ("late-talker syndrome"), his symptomology at a young age was atypical of a late talker. Infants and toddlers with later-diagnosed CAS are found to present differently between birth and 24 months from those with typical speech sound production. Differences are evident in volubility (the quality of talking fluently), phonetic diversity, and syllable shapes.[17] Our patient experienced several of these behaviors, including absent cooing and babbling, as well as a limitation in stimulable consonants and vowels.

Therapy was completed in both center-based (45 minutes 1 to 2 times per week) as well as home-based environments (60 minutes 1 time per week). These services were coordinated through therapist communication and observation as well as sharing of goals. Initial therapy sessions were focused on oro-motor control and imitation, as well as repetition of mouth movements. Sarah Rosenfeld Johnson's program of whistles and straws (www.talktools.com) was used to improve oral motor control for expanding airflow. Sound approximations for words with previously established signs were the next goals of the therapy sessions. The Kaufman Speech Praxis Program was used in recommended progression from simple sound/syllable combinations to more complex sound and syllable productions. Success was expected at each level before advancing. Visual models and intermittent touch cues were used to cue place of articulation (such as tap to lips for bilabials [m,p,b,w], tap to throat for back sounds). This method was used consistently at each session to improve approximations. He underwent high-frequency treatment twice a week with additional once-weekly at-home speech therapy. His first notable improvement was in oro-motor control evidenced by decreased drooling. The patient's mother was educated and provided a home program to facilitate increased tongue mobility via food-based oral stimulation and lingual movement. In the home environment, they continued to use sign language with speech prompting. Subsequently, as he developed new words, he replaced the previous signed communication for natural speech. Mother and speech language pathologist reported during an interview that sign language helped to prevent and dissipate frustration when verbal communication broke down, aided in day-to-day activities, and facilitated language acquisition. As clarity and spontaneity progressed throughout therapy, the patient became more social and engaging, with vastly improving play and interaction skills with peers and adults.

As shown in Table 2, the patient had mildly delayed expressive language at 1 year 10 months of age with noted improvement over 2 years of therapy with normal receptive language throughout. He was unable to have age-normative values for the Kaufman Speech Praxis Test at diagnosis due to his young age, but his observational findings were consistent with significant speech motor planning deficits. Still notable at age 3 was difficulty in speech motor planning for longer and more complex utterances. His testing after completion of therapy showed marked improvement and near resolution of previously described deficits (Table 3). The Verbal Motor Production Assessment for Children was used at 3 years 9 months of age and was within age-appropriate scoring in all areas, including general

TABLE 2 Language Testing

Test Name	Age	Standard Scores
Preschool Language Scale 5	1 y 10 mo	Auditory Comprehension: 120 Expressive Language: 85
Preschool Language Scale 5	3 y	Auditory Comprehension: 104 Expressive Language: 90
Clinical Evaluation of Language Fundamentals Preschool 2	3 y 6 mo	Core Language Index: 102 Receptive Language Index: 105 Expressive Language Index: 104 Language Content Index: 102 Language Structure Index: 110

motor control (100%), focal oro-motor control (85%), sequencing (76%), connected speech and language control (87%), and speech characteristics (100%). Cumulatively, his testing after completion of therapy was largely within age-appropriate normative values and showed gains in areas of speech motor planning and speech and language control and expression.

At his final speech therapy appointment at age 3 years 6 months, our patient was noted with the following age-acceptable speech sound substitutions: k/tr, b/sp, s/z, b/v, s/sw, s/sh, k/ch, f/th, g/dz, f/fl, g/dr, f/fr, kw/kl. These patterns of consonant cluster simplifications are judged age appropriate. His error substitution for /v/ is common and stimulable. Other errors documented previously were not remediable at this time, do not affect overall intelligibility, and will be monitored as his speech continues to develop. Although not formally assessed with a polysyllabic test or non–word-repetition test, no evidence of prosody or resonance deficits were noted at his current language/speech level.

When comparing our patient's speech with pretreatment levels, the following was noted. Before addition of our formal speech therapy program, our patient had no functional verbal communication. After treatment he was communicating with ease to familiar partners at a conversational level. Parents reported understanding 90% to 100% of what he said and the speech therapist was able to comprehend >80% of communication on documented conversation sample analysis. In addition, phrase length increased from single syllables, early into his therapy program, to 8+ syllable phrases that included accurate use of auxiliary and verb endings as well as 3- to 4-syllable word productions. He also rarely used sign language.

DISCUSSION

Our patient showed significant gains by using sign language while participating in an intensive speech motor planning program. Our patient's multimodal approach to treatment reduced frustration with communication and served as a method for development of both receptive and expressive language. He was able to develop and expand on social pragmatic skills with both peers and adults. His success was also likely associated with the high frequency of motor-based speech therapy at an early age and dedication to his home program. We suspect that early introduction of sign language by family proved to be a highly effective form of language development that when coupled with intensive oro-motor and speech sound therapy resulted in rapid resolution of symptoms. As his language developed, he independently discontinued the use of sign language, which provides evidence to assuage parental concerns that sign language inhibits speech development.

Successful intervention requires skilled speech-language pathologists to manage therapy to improve linguistics and speech motor planning while developing language and communication skills. Although speech development in children with CAS can be slow, AAC strategies can facilitate improvement in both receptive and expressive language skills without the challenges of

TABLE 3 Kaufman Speech Praxis Test

	Observations at 18 mo	Standard Scores at Age 3 y	Standard Scores at Age 3 y 9 mo (After Completion of Therapy)
Part 1: Oral movement	• No tongue lateralization • No lip protrusion • Marked drooling	114	110
Part 2: Simple phonemic/syllabic level	• Three vowel approximations • Three syllable approximations	103	110
Part 3: Complex phonemic/syllabic level	• None	91	91
Part 4: Spontaneous length and complexity	• None	80	87

natural speech production.[7] Cumley and Swanson[6] and King et al[18] observed improved speech and functional communication, including natural speech, when AAC was used in a multimodal intervention. These same findings were echoed in the qualitative reports of our patient's mother and his speech language pathologist. Despite the success of sign language, it should not be used solely but rather coupled with verbal attempts.[15]

It is well known that there is a paucity of published literature on many commonly used treatment methods for CAS; however, our case suggests that using a multimodal approach to children with nonspeech oral apraxia and CAS may have additive benefits. Our patient received sign language augmentation, the Sarah Rosenfeld Johnson program of whistles and straws, and the Kaufman Speech Praxis Program. All interventions have little to no published evidence and further research is certainly indicated to help guide best practice. This case report provides some preliminary evidence for all of them used in combination.

Our patient and his success reiterate that sign language should be considered within a multimodal approach with intensive speech therapy for children with CAS.

Limitations to the use of sign language include motor planning deficits in fine motor or gross motor skills or lack of interest in using sign. Tukel et al[11] showed a relationship between CAS and heterogeneous motor problems suggestive of a global motor delay. This delay makes the precise motor movements of the hands and arms of sign language more difficult to understand. In these children, a different AAC may be more appropriate.[6] Sign may not be indicated for children with associated speech motor planning difficulties, but it could be considered as a method for visual cueing between parent and child, where the parent signing creates an association with the visual cue and the word/sound.[15] Decision-making for intervention strategies ultimately depend on the communication need of the client and should be driven by assessment findings.[7]

CONCLUSIONS

In our patient, the diagnosis of CAS was highly suspected due to non–speech-related oro-motor dysfunction, evidence of motor groping, and a strong desire and effort to communicate without successful sound production. Although the rapidity of our patient's speech improvement before the age of 3 may have suggested constitutional speech delay, his symptomology at a young age was atypical of a late talker. We suspect that early introduction of sign language by family, even before the patient was suspected or confirmed to have CAS, proved to be a highly effective form of language development that when coupled with an intensive oro-motor and speech sound therapy program resulted in rapid resolution of symptoms.

As his speech clarity improved, he independently discontinued the use of sign language, which provides evidence to assuage parental concerns that sign language inhibits speech development. We agree that this child may have been exceptional in many respects. Further research is needed to determine to what extent the use of sign language accounted for his rapid and nearly complete resolution of CAS. We caution parents and physicians that not all cases of CAS will respond in a similar fashion, but hope that research will help identify a subset in whom this outcome may be more readily achieved.

The child's parents signed consent and were given the opportunity to review this case report before submission.

ACKNOWLEDGMENTS

We thank Ms Sharon Gretz, Executive Director at Childhood Apraxia of Speech Association of North America for her insightful suggestions and edits to our paper.

ABBREVIATIONS

AAC: augmentative and alternative communication
CAS: childhood apraxia of speech

Development, Section Chief Behavior and Developmental Pediatrics, Penn State Hershey Children's Hospital, 500 University Dr, H085, Hershey, PA 17033-0850.
E-mail: ctierney@hmc.psu.edu

PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

FINANCIAL DISCLOSURE: The authors have indicated they have no financial relationships relevant to this article to disclose.

FUNDING: No external funding.

POTENTIAL CONFLICT OF INTEREST: The authors have indicated they have no potential conflicts of interest to disclose.

REFERENCES

1. Ad Hoc Committee on Childhood Apraxia of Speech; American Speech-Language-Hearing Association. Position statement: Childhood apraxia of speech. 2007. Available at: http://www.asha.org/policy/ps2007-00277.htm. Accessed April 23, 2015
2. Grizzle KL, Simms MD. Language and learning: a discussion of typical and disordered development. *Curr Probl Pediatr Adolesc Health Care*. 2009;39(7):168–189 [Review]
3. Namasivayam AK, Pukonen M, Goshulak D, et al. Treatment intensity and childhood apraxia of speech. *Int J Lang Commun Disord*. 2015;50(4):529–546
4. Murray E, McCabe P, Ballard KJ. A comparison of two treatments for childhood apraxia of speech: methods and treatment protocol for a parallel group randomised control trial. *BMC Pediatr*. 2012;12(1):112
5. Murray E, McCabe P, Ballard KJ. A systematic review of treatment outcomes for children with childhood apraxia of speech. *Am J Speech Lang Pathol*. 2014;23(3):486–504
6. Cumley G, Swanson S. Augmentative and alternative communication options for children with developmental apraxia of speech: three case studies. *Augment Altern Commun*. 1999;15(2):110–125
7. Oommen ER, McCarthy JW. Simultaneous natural speech and AAC interventions for children with childhood apraxia of speech: lessons from a speech-language pathologist focus group. *Augment Altern Commun*. 2015;31(1):63–76
8. Martikainen A, Korpilahti P. Intervention for childhood apraxia of speech: A Single-case study. *Child Lang Teach Ther*. 2011;27(1):9–20
9. Luke C. Impact of speech-generating devices on the language development of a child with childhood apraxia of speech: a case study. *Disabil Rehabil Assist Technol*. 2016;11(1):80–88
10. Bornman J, Alant E, Meiring E. The use of a digital voice output device to facilitate language development in a child with developmental apraxia of speech: a case study. *Disabil Rehabil*. 2001;23(14):623–634
11. Tukel S, Bjorelius H, Henningsson G, McAllister A, Eliasson AC. Motor functions and adaptive behaviour in children with childhood apraxia of speech. *Int J Speech Lang Pathol*. 2015;17(5):470–480
12. Maas E, Gildersleeve-Neumann C, Jakielski KJ, Stoeckel R. Motor-based intervention protocols in treatment of childhood apraxia of speech (CAS). *Curr Dev Disord Rep*. 2014;1(3):197–206
13. Murray E, McCabe P, Ballard KJ. A randomized controlled trial for children with childhood apraxia of speech comparing rapid syllable transition treatment and the Nuffield Dyspraxia Programme-third edition. *J Speech Lang Hear Res*. 2015;58(3):669–686
14. Glennen SL, DeCoste DC. *The Handbook of Augmentative and Alternative Communication*. San Diego, CA: Singular Publishing Group; 1997
15. Gretz S. Using sign language with children who have apraxia of speech. Apraxia-KIDS: a program of The Childhood Apraxia of Speech Association (CASANA). Available at: www.apraxia-kids.org/library/using-sign-language-with-children-who-have-apraxia-of-speech/. Accessed April 23, 2015
16. Hall PK, Jordan LS, Robin DA. *Developmental Apraxia of Speech: Theory and Clinical Practice*. Austin, TX: Pro-Ed; 1993
17. Overby M, Caspari SS. Volubility, consonant, and syllable characteristics in infants and toddlers later diagnosed with childhood apraxia of speech: a pilot study. *J Commun Disord*. 2015;55:44–62
18. King AM, Hengst JA, DeThorne LS. Severe speech sound disorders: an integrated multimodal intervention. *Lang Speech Hear Serv Sch*. 2013;44(2):195–210

Bridging the Gap Between Speech and Language: Using Multimodal Treatment in a Child With Apraxia

Cheryl D. Tierney, Kathleen Pitterle, Marie Kurtz, Mark Nakhla and Carlyn Todorow

Pediatrics 2016;138;
DOI: 10.1542/peds.2016-0007 originally published online August 4, 2016;

PEDIATRICS®
OFFICIAL JOURNAL OF THE AMERICAN ACADEMY OF PEDIATRICS

Bridging the Gap Between Speech and Language: Using Multimodal Treatment in a Child With Apraxia

Cheryl D. Tierney, Kathleen Pitterle, Marie Kurtz, Mark Nakhla and Carlyn Todorow

Pediatrics 2016;138;
DOI: 10.1542/peds.2016-0007 originally published online August 4, 2016;