Speech Pathology
Sydney School of Health Sciences
The University of Sydney Australia
ABN 15 211 513 464 CRICOS 00026A
E tricia.mccabe@sydney.edu.au
https://www.sydney.edu.au/medicine-health/about/our-people/academic-staff/tricia.mccabe.html

Evidence Summary - Childhood Apraxia of Speech – January 2024

McCabe, P., Murray, E. & Thomas, D.*

This document is a free summary of the current evidence on assessment, diagnosis and treatment of Childhood Apraxia of Speech (CAS; aka Dyspraxia). Please seek advice from your speech pathologist.

This evidence summary is only valid until December 2025.

Background

Childhood Apraxia of Speech is a severe, permanent, and lifelong neurodevelopmental disorder of speech motor programming and planning which is present from birth and does not naturally resolve. In recent years, substantial progress has been made in improving speech pathology treatment for CAS but there remains a large number of older children, adolescents and adults who have severe limitations to all aspects of their lives due to ineffective and/or insufficient treatment in earlier years. Recent advances in treatment efficacy in preschool and primary years should reduce this extended prevalence tail over time however there is emerging evidence that a significant burden of psychosocial, educational, economic and communication deficits remains across the lifespan with resultant restrictions on participation and daily life.

Most people with CAS have an unknown cause, however CAS can co-occur with all other developmental conditions including other communication disorders. As many as 1 in 3 children have a genetic cause for their CAS, whether it is a single gene change or a chromosomal deletion or duplication. CAS also has increased frequency in children and adults with rare single gene conditions (e.g., *SETBP1-HD, FOXP2*), galactosemia, intellectual disability, global developmental delay, epilepsy, and some minimally verbal autistic people. However, it appears to have no increased prevalence in verbal autistic children above the population prevalence of approximately 1 in 1000 children.

CAS appears to exist in most, if not all, languages and has been documented in at least Arabic, Cantonese, Danish, Dutch, French, German, Italian, Portuguese, Spanish, Swedish, Tagalog, Turkish and English.

Assessment

Diagnosis of CAS requires skilled assessment by **a suitably qualified and experienced speech pathologist**. Best practice in assessment depends on the child's age, severity and comorbidities and should use at least 3 speech tasks. Additional tasks are required to describe other parts of communication, such as language, reading, spelling or social communication.

Suggested Assessment Protocols in English*

*** Assessment of CAS in languages other than English will require similar tasks however the literature is less clear about which to select.**

Younger children or those with severe speech difficulties	**Older children or those with milder speech difficulties**
Hearing screening or assessment	
Comprehensive oral musculature structural and functional evaluation	
Single word productions appropriate for age, language, accent, and culture	
Single word list should ideally include 50 common words with a range of sounds, syllable shapes and number, and word shapes.	Single word list should include at least 30 polysyllabic words including weak onset word structures. Single and two syllable words can be included.
Specific tasks can include: • Diagnostic Evaluation of Motor Speech Skills (DEMSS, Strand & McCauley, 2019) OR • Nuffield Dyspraxia Programme (NDP3) Assessment (Williams & Stephens, 2009) OR • Verbal Motor Production Assessment for Children-Revised (VMPAC-R, Hayden & Namasivayam, 2021). • Diagnostic Evaluation of Articulation and Phonology – Inconsistency Subtest (Dodd et al, 2002)	Specific tasks could include: • Speech diadochokinesis tasks (e.g. 'peteke') (Diepeveen et al, 2019) • Syllable Repetition Test (Shriberg et al, 2012) • Multisyllabic Rapid Naming tasks (Preston et al, 2021) • "Buy Bobby A Puppy" inconsistency assessment (Iuzzini-Seigel et al, 2017)
Connected speech samples	
Sample of typical communication in interaction with their carer or in play (including use of sounds, speech, gesture and communication devices). It could include a picture description or talking about a favourite toy or photo.	Sample of connected speech including polysyllabic words (words of 3 or more syllables) to sample the child's connected speech accuracy and prosody. This could include a story retell, conversation or a polysyllablic words in sentences task.

Diagnosis of CAS requires that a child at a minimum meets all three ASHA (2007) consensus-based features of CAS:

1. *Inconsistency across words and syllables*
2. *Lengthened and disrupted coarticulatory transitions.*
3. *Inappropriate prosody.*

CAS can also be diagnosed if a person has vowel distortions and at least another 3 /10 criteria on the Mayo 10+1 checklist (Shriberg et al, 2012, Table 1). Iuzzini-Seigel et al, (2022) developed a tool which may be used discriminate CAS from dysarthria. **Please note:** for a diagnosis of CAS to be accurate, children need to have a clear intent to communicate regardless of age or severity. Slow progress in speech therapy is not diagnostic of CAS.

Severity of CAS has not been formally defined however clinicians may use the following factors in determining severity:

1. Intelligibility – children with more severe CAS will struggle to be intelligible even to immediate family.
2. Speech inventory (number of sounds and syllable structures) in comparison to other people of the same chronological or language age.
3. Number of features of CAS present and severity of features. These lists of features come from two sources (ASHA, 2007 and Shriberg et al, 2012).
4. In older children, adolescents, and adults: Difficulty saying new or longer words, avoiding speaking tasks such as using the phone, social isolation, or reduced quality of life.
5. Presence of other communication or cognitive issues.

Treatment

The first randomised controlled trial comparing two CAS treatments was published in 2015. Murray, McCabe and Ballard (2015) compared the Nuffield Dyspraxia Programme (3rd ed; NDP3) with Rapid Syllable Transition Treatment (ReST). Both treatments were effective in changing the speech of children aged 4-12 with CAS. NDP3 had better immediate effect and ReST had a better long-term effect. Both treatments are therefore currently recommended when delivered as per the RCT (ie 4 days per week for 3 weeks @ 1 hour per day).

The NDP3 was also tested in a study comparing two versions (McKechnie et al., 2020) and ReST has been compared with Ultrasound Biofeedback (McCabe et al., 2023). There was no difference between ReST and ultrasound when therapy was twice per week for 6 weeks. Both studies may have shown differences if more children had been included. Work is underway on RCTs evaluating other CAS treatments and comparing therapy to no therapy.

Three systematic reviews have been conducted in the past 10 years. The first two (Murray et al., 2014; Maas et al., 2014) examined a broad range of treatment evidence with a range of quality. Murray et al (2014) recommended:

1. **Rapid Syllable Transition Treatment (ReST)**
2. **Nuffield Dyspraxia Programme 3rd edition (NDP3)**
3. **Dynamic Temporal and Tactile Cueing (DTTC)**
4. **Integrated Phonological Awareness (IPA)**

From this list, ReST and IPA are suitable for children with less severe difficulties and/or older children. DTTC and NDP3 are more suitable for children with more severe difficulties and/ or younger children. Resources and training for ReST, IPA and DTTC are freely available on the internet. The NDP3 has a manual with stimuli kits and training which can be purchased online.

In addition to the treatments listed above, Maas et al., (2014) also included:

5. **Ultrasound biofeedback**

This is more suitable for primary school aged children and older with milder speech issues. Ultrasound biofeedback is beyond the scope of many clinicians due to cost of equipment.

In the most recent systematic review, Morgan et al., (2018) in the Cochrane Database reported that only ReST and NDP3 had RCT level evidence and called for more treatment research. There is limited research to guide treatment decisions when children have more than one co-occurring speech disorder, however, a logical choice would be to select a treatment that has demonstrated effectiveness for both of the child's speech disorders. For example, a child with dysarthria and CAS may benefit from DTTC or ReST which have evidence of efficacy with both disorders.

Other treatments have less well-developed evidence and should be undertaken with caution as they have not yet been shown to be effective in multiple and/or randomised studies of children who clearly had CAS.

Treatment intensity and frequency

On average effective treatment requires 2-6 sessions per week for an undescribed maximum (more than 1 year) (Maas et al., 2014). The CAS treatment evidence shows that therapy 4 times a week in blocks of 12-16 sessions followed by a 4-6 week break from therapy with repeated cycles of therapy is optimal (Murray et al, 2015). All studies have shown that the greater the treatment intensity, the more effective the therapy, and the more efficient the progress (e.g., Edeal and

Gildersleeve-Neumann, 2012). Session length ideally should be 45-60 minutes but will depend on both the child and the treatment selected. Two studies have explicitly tested therapy once per week and shown it to be ineffective (Namasivayam et al, 2015; Thomas et al., 2023).

Group Therapy

There is no evidence for any group treatment being trialed in any level of research with anyone with CAS since 1960. Group treatment is not recommended for any CAS feature and there is no theoretically sound reason for it to be trialed. People with CAS may benefit from evidence-based group therapy interventions for their co-morbid conditions but again there is no research evidence for such treatments in people with CAS who have these comorbid conditions.

Therapy by people who are not Speech Pathologists.

There is very limited evidence that therapy for CAS can be provided by anyone other than a speech pathologist. In all but three studies, speech pathologists or supervised speech pathology students have provided therapy. Two studies (Thomas et al., 2017; Lim et al., 2020) trialed parent delivered therapy with limited success and it is not currently recommended. One study (Lim et al, 2019) trained teacher's aides providing DTTC therapy which was moderately successful.

References

American Speech-Language-Hearing Association. (2007). Childhood Apraxia of Speech [Technical Report]. www.asha.org/policy.

Diepeveen, S., van Haaften, L., Terband, H., de Swart, B., & Maassen, B. (2019). A Standardized Protocol for Maximum Repetition Rate Assessment in Children. *Folia phoniatrica et logopaedica*: 71(5-6), 238–250.

Dodd, B., Hua, Z., Crosbie, S., Holm, A., & Ozanne, A. (2002). *Diagnostic evaluation of articulation and phonology (DEAP).* London, England: The Psych Corp.

Edeal, D. M., & Gildersleeve-Neumann, C. E. (2011). The importance of production frequency in therapy for childhood apraxia of speech. *AJSLP, 20*(2), 95

Eising E, Carrion-Castillo A, Vino A, Strand EA, Jakielski KJ, Scerri TS et al. A set of regulatory genes co-expressed in embryonic human brain is implicated in disrupted speech development. Mol Psychiatry 2019; 24(7): 1065-78.

Hayden, D., & Namasivayam, A., (2021) *Verbal Motor production Assessment for children – Revised edition*. Available from: https://vmpac-r.com/

Iuzzini-Seigel, J., Allison, K. M., & Stoeckel, R. (2022). A tool for differential diagnosis of childhood apraxia of speech and dysarthria in children: A Tutorial. *LSHSS:* 53(4), 926–946.

Hildebrand, M.S., Jackson, V.E., Scerri, T.S., Van Reyk, O., Coleman, M., Braden, R.O. et al. (2020). Severe childhood speech disorder: Gene discovery highlights transcriptional dysregulation. *Neurology*; 94(20): e2148-e67.

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.

Kaspi, A., Hildebrand, M.S., Jackson, V.E., Braden, R., van Reyk, O., Howell, T. et al. (2023). Genetic aetiologies for childhood speech disorder: novel pathways co-expressed during brain development, *Molecular Psychiatry; 28*: 1647–1663.

Lim, J., McCabe, P., & Purcell, A. (2019). Another tool in my toolbox": Training school teaching assistants to use Dynamic Temporal and Tactile Cueing with children with CAS. *Child Lang Teaching & Therapy. 35* (3) 241–256.

Lim, J. M., McCabe, P., & Purcell, A. (2020). Look at Mummy: challenges in training parents to deliver a home treatment program for childhood apraxia of speech in a rural Canadian community. *Rural and Remote Health, 20*(2), 59-68.

Maas, E., Gildersleeve-Neumann, C. E., Jakielski, K. J., & Stoeckel, R. (2014). Motor-based intervention protocols in treatment of childhood apraxia of speech (CAS). *Current Developmental Disorders Reports, 1* 197.

McCabe, P., Preston, J. L., Evans, P., & Heard, R. (2023). A pilot randomized control trial of motor-based treatments for childhood apraxia of speech: Rapid Syllable Transition Treatment and Ultrasound Biofeedback. *AJSLP, 32*(2), 629

McKechnie, J.,…, Murray, E., McCabe, P., & Ballard, K. J. (2020). The influence of type of feedback during tablet-based delivery of intensive treatment for childhood apraxia of speech. *JCD: 87*, 106026.

Morgan, A. T., Murray, E., & Liégeois, F. J. (2018). Interventions for childhood apraxia of speech. *Cochrane Datab of Syst Rev, 2018* (5).

Morgan, A.T., Amor, D., St John, M., Scheffer, I,E., Hildebrand, M. (in press). Genetic architecture of childhood speech disorder: a review. Molecular Psychiatry.

Murray, E., McCabe, P. & Ballard, K.J. (2015). A randomized control trial of treatments for childhood apraxia of speech. *JSLHR: 58,* (3) 669-686.

Murray, E., McCabe, P. Heard, R. & Ballard, K.J. (2015). Differential diagnosis of children with suspected childhood apraxia of speech. *JSLHR: 58*,(1) 43-60.

Murray, E., McCabe, P., & Ballard, K. J. (2014). A systematic review of treatment outcomes for children with childhood apraxia of speech. *AJSLP: 23*(3), 486-504.

Namasivayam, A. K., Pukonen, M., Goshulak, D., Hard, J., Rudzicz, F& ... Lieshout, P. (2015). Treatment intensity and childhood apraxia of speech. *IJLCD: 50*(4), 529-546.

Preston, J.L., Brick, N., & Landi, N. (2013). Ultrasound biofeedback treatment for persisting childhood apraxia of speech. *AJSLP. 22* 627–43.

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.

Strand, E. A., & McCauley, R. J. (2019). *Dynamic Evaluation of Motor Speech Skill (DEMSS) Manual*. Baltimore, MD: Brookes Publishing.

Thomas, D. C., McCabe, P., & Ballard, K. J. (2017). Combined clinician-parent delivery of rapid syllable transition (ReST) treatment for childhood apraxia of speech. *IJSLP:* 20(7), 683-698.

Thomas, D. C., McCabe, P., Ballard, K. J., & Bricker-Katz, G. (2018). Parent experiences of variations in service delivery of …ReST treatment for childhood apraxia of speech. *Developmental Neurorehab., 21*(6), 391-401.

Thomas, D. C., McCabe, P., Ballard, K. J., & Lincoln, M. (2016). Telehealth delivery of Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech. *IJLCD: 51*(6), 654-671.

Thomas, D., Murray, E., Williamson, E., & McCabe, P. (2023). Weekly treatment for childhood apraxia of speech with Rapid Syllable Transition Treatment: A single-case experimental design study. *JSLHR:* 1-22.

Williams & Stephens (2009). *The Nuffield Dyspraxia Programme.* Retrieved from http://www.ndp3.org

* Thanks to Professor Angela Morgan for her comments on the causes section of this document.

Contact: tricia.mccabe@sydney.edu.au for more information