Childhood Apraxia of Speech: Recent Research to Inform Best Practice

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Disclosures

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Plan for Today

Review findings of recent research that contribute to our understanding of CAS as a motor speech disorder

- Assessment: identify features and tasks that contribute to diagnosis of CAS
- Treatment: inform decision-making for treatment of CAS, with an emphasis on Principles of Motor Learning
- CAS+: Briefly highlight recent articles on associated issues/diagnoses among children with CAS

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Introduction

1954: Muriel Morley and colleagues provided an early description of motor-based speech disorders in children

1974: Kathe Yoss and Fred Darley used the term "apraxia" to describe children exhibiting characteristics similar to adults with apraxia

2007: The ASHA Technical Report was released, providing a review of progress in identifying CAS as a unique SSD

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Introduction

The ASHA Technical spurred efforts by researchers to…

- analyze speech characteristics to identify how CAS is unique from other SSDs,
- develop tasks and tools for differential diagnosis,

and

- examine effectiveness of interventions

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Assessment

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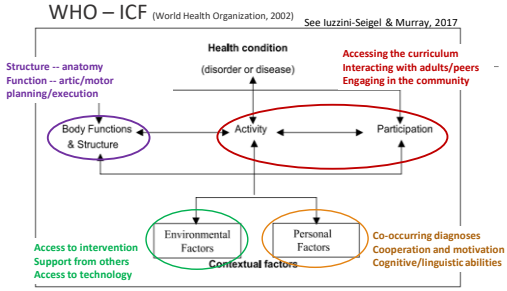
WHO – ICF (World Health Organization, 2002)

See Iuzzini-Seigel & Murray, 2017



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Communicative intent

Express meaning/word retrieval
Map sound patterns/phonology
Grammar/syntax
Prosody/Pragmatics

Movement parameters
Direction, Speed, ROM,Force

Produce phonemes & syllables
Respiration
Phonation
Articulation
Resonance

Cognitive

Linguistic

Motor Planning/Programming

Motor Execution

Adapted from a model by Edythe Strand

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Assessment

Develop an individualized assessment plan:

- ❑ History
- ❑ Oral structure/function exam
- ❑ Speech, language sampling
- ❑ Standardized speech/language testing
- ❑ Assess motor speech skill
- ❑ Assess phonological awareness/literacy as appropriate

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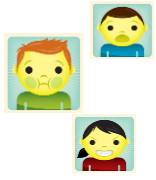
Oral Structure & Function

- Structures
  - Range of motion
  - Coordination
  - Strength
  - Ability to vary muscular tension
  - Speed
- Tissue characteristics



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Oral Structure & Function

Hypotonia ≠ weakness

- Hypotonia: reduced muscle activation *at rest* (not during speech)
- Weakness: reduced strength
  - Strength: muscle contraction, causing *movement* of a structure (e.g. during speech)

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Oral Structure & Function

- There appears to be a strong association between lip and jaw movements and measures of cognition and language
  - maintained even when accounting for age (Nip, Green, & Marx 2010)
- A wide range of variability was found among typical speakers -- from children to adults -- in an alternating tongue lateralization task (Small, McAllister, & Grigos, 2018)
- Tongue strength does not appear to be related to severity of speech sound disorder (Potter, Nievergelt & VanDam, 2019)

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Take-Home Message

- Consider a child's developmental level when assessing speech motor skills
- Assessment of muscles in action is required to establish presence or absence of weakness
- Make observations of articulator function but be wary of over-interpretation

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Standardized Speech/Language Testing: Language Sampling

- Language sampling is possible with children who have severe speech impairments (Binger, Ragsdale, & Bustos, 2016)
- Children with CAS have been found to exhibit errors in expressive morphosyntax that are not easily explained by their speech errors (McNeill & Gillon, 2013; Murray, Thomas, & McKechne, 2019)

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Standardized Speech/LanguageTesting: Language Sampling

- Examining the child's verbal output relative to language skill can inform diagnosis
  - CAS diagnosis does NOT require a receptive > expressive discrepancy, but studies suggest likelihood of unbalanced skills (e.g., McNeill & Gillon, 2013; Murray, Thomas, & McKechnie, 2019)

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Standardized Speech/Language Testing: Speech Sampling

- Analysis of phonetic and phonemic inventories will help to clarify patterns of error that may be missed if only standardized testing is used.
  - Can also inform target selection
- Consider speech skill discrepancies between single word tasks and contextualized language – varied task complexity (Iuzzini-Seigel, et al., 2017; Murray, et al., 2015; Strand, et al., 2013)
- A connected speech protocol has been suggested for children with CAS (Barrett et al., 2020)

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Take-Home Message

- Differential diagnosis of CAS involves consideration of factors and skills other than motor speech skill
  (this is probably not news to anyone)

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Assessment

Develop an individualized assessment plan:

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Assessment of Motor Speech Skill: Tests and Tasks

No single test is adequate; a combination of measures is recommended (Allison et al., 2020; Murray et al., 2020; Strand & McCauley, 2008)

Some measures that have been used clinically and in research include:

- Multisyllable Word Repetition (e.g., Benway & Preston, 2020; Murray et al., 2015)
- Maximum Performance Tasks (e.g., Diepeven et al., 2019; Rvachew et al., 2005)
- Syllable Repetition Task (Rvachew & Matthews, 2017;Shriberg et al., 2012)
- Pause Marker (Shriberg et al., 2017a)

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Motor Speech Skills

Movement specifications include:
- range of motion (/a/ vs /i/)
- direction of movement (/ai/ vs /ja/
- speed of movement
- force/muscle tension (/m/ vs /b/)

Additionally influenced by:
- pitch, duration, loudness
- initial and final articulatory configuration is bidirectional – influenced by coarticulatory effects

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Discriminative Features: Scoping Review (Allison, et al., 2020)

- More consistent protocols and data collection techniques are needed.
- Consensus is building around use of operationalized feature sets:
  - Characteristics contained in the ASHA Technical Report on CAS and
  - Mayo 10 criteria (Shriberg et al., 2011)
    - (e.g., any 4 of 10 features over at least 3 assessment tasks)

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Discriminative Features: Scoping Review

- Most studies using surface speech characteristics suggested these features:
  - Reduced segmental accuracy and/or greater error inconsistency (Aziz et al., 2010; Iuzzini-Seigel et al., 2017; Murray et al., 2015)
  - Prosodic deficits or lexical stress errors (Aziz et al., 2010;Iuzzini-Seigel, et al., 2017;Murray, et al., 2105)
  - With task complexity a factor in performance (Iuzzini-Seigel, et al., 2017; Murray, et al., 2015; Strand, et al., 2013)

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Discriminative Features: Research Evidence

- Children with CAS have
  - Reduced accuracy, longer movement duration, and greater speech motor variability than TD peers
  - Reduced accuracy and greater articulator movement variability than children with other SSDs

(Case & Grigos, 2020; Case & Grigos, 2016; Grigos, 2016; Grigos, Moss, & Lu, 2015; Moss & Grigos, 2012; Terband, et al.,2011)

- Pause Marker may be a useful quantitative measure

(Allison et al., 2020; Shriberg et al., 2017; Tilkens et al., 2017)

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Differential Diagnosis: Systematic Review (Murray, et al., 2020)

- 15 studies were reviewed based on contribution to efforts to support differential diagnosis
  - Studies included perceptual ratings, acoustic measures, and kinematic markers
- No study met highest level of study quality
- No consistent reference test used across studies
- Some "promising" tools identified

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Systematic Review: *Possible* Protocol

Young child

Dynamic Evaluation of Motor Speech Skill (DEMSS)	Strand & McCauley, 2019
Iuzzini-Seigel inconsistency measure	Iuzzini-Seigel et al., 2017a
And	
Robbins & Klee oral musculature assessment }	Murray, McCabe, & Ballard 2015
Polysyllable test }	
Or	
Maximum Performance Task	Diepeven et al., 2019; Rvachew et al., 2005

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Systematic Review: *Possible* Protocols

CAS vs SD

(Oral musculature assessment)

Pause Marker — Tilkens et al.,2017 protocol

and/or

Inconsistency measure — Iuzzini-Seigel et al., 2017a

CAS vs dysarthria

Thorough oral musculature assessment } Murray, McCabe, & Ballard 2015

Polysyllable task }

Maximum Performance Tasks — Diepeven et al., 2019; Rvachew et al., 2005

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Special Case: Early Diagnosis

Retrospective analyses of home videos suggests that consonant development of children between birth and age 3 years who were later diagnosed with CAS may be different than TD children or children with other SSDs

- Differences include…
  - less vocalizations overall,
  - fewer consonants,
  - less diverse phonetic repertoire, and
  - later consonant acquisition

(Overby, Belardi, & Schreiber, 2019; Overby & Caspari, 2015 Overby, Caspari, & Schreiber, 2019)

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Special Case: Early Diagnosis

Given the need for additional research, *provisional diagnosis* is suggested for children below age 3 years:

"CAS cannot be ruled out,"

"Signs are consistent with problems in planning the movements required for speech,"

"Suspected CAS."

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Take-Home Message

- Combinations of measures are needed, given the limited, but growing, evidence for sufficiently sensitive and specific individual markers
  - Diagnosis is not dependent on age or number of words
  - Speech skills should be sampled in a range of complexity
- Clinical practice: SLPs should choose a study or studies in which the children have similar characteristics to their student/client

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Intervention

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Intervention

- Intensive treatment is recommended (e.g., ASHA 2007; Namasivayam et al., 2015)
- Response to treatment may vary due to both intrinsic and extrinsic factors
  - Children present with a range of severity of their speech motor impairment, and a variety of co-occurring/comorbid conditions
- Intervention will need to be modified based on a child's progress or lack of progress

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Intervention

- Consider factors on the ICF model when planning goals (Iuzzini & Murray 2017)
  - Rusiewicz et al., (2018) reported parents' concern that CAS affected everyday activities and social interactions
- There is no identified intervention approach that has been shown to be effective for all children with CAS or for every stage of therapy
  - But we are able to identify important ingredients in effective therapy

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#1 Ingredient: Motor Based Intervention to Improve MOVEMENT

Paradigm shift: sounds → MOVEMENTS

Address the inefficiency in specifying parameters of movement

Movements need to occur

- at the right time
- in the right direction
- with the right force

to get to

- the right place
- at the right time

Thanks to Edwin Maas for sharing material used in this section

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#1 Ingredient: Motor Based Intervention

- Intervention should facilitate motor LEARNING, not just motor PERFORMANCE
  - Including retention (short term change) and transfer/generalization (longer term, beyond movement-specific)
  - Achieving accuracy in therapy/practice does not equal learning to use the skill in other contexts/conditions

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Other Important Ingredients (Maas, et al., 2008)

Understanding the task: To improve *movement vs* sounds

- Are the strategies appropriate for the child's developmental ability and motor skill?
- Use Pre-practice
  - Explain the task and relevance
  - Demonstrate how you will work
  - Explain/demonstrate accurate response
  - Briefly practice with 100% feedback

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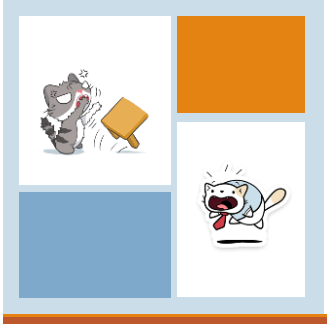

Other Important Ingredients

Stimulability

- If the child is not stimulable, the result may be frustration and distrust

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Other Important Ingredients (Strand & Debertine, 2000)

Functional Targets

- Think about the needs of the "whole child"
- Build vocabulary and language as well as speech accuracy
- Give the child ways to interact with others and with their environment
- Based on the child's phonetic/phonemic inventory
  - and ongoing efforts to expand inventory

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Motor Based Intervention

Where do I start/what position? Where do I stop?

How do I get there (motor commands)?

What did it sound like?

How does it feel when I do it accurately (proprioception/sensory)?

Can I do it again in a new context?

Time to talk about Challenge Point Framework and Principles of Motor Learning (PML)

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Intervention: Challenge Point Framework

(Guadagnoli and Lee, 2004)

Learning a given task will depend on balancing

- the skill level of the child
- task complexity
- context

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Intervention: Challenge Point Framework

An optimal challenge point maximizes learning and minimizes negative practice

- This may require manipulation of different aspects of the principles of motor learning (PML) at different times during treatment

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Intervention: Motor Learning & PML

- These principles have been shown to facilitate motor learning in research on limbs.
- There is less research for speech or for CAS in particular (Maas, et al., 2008)
- A number of studies have attempted to assess contribution of specific principles (Maas, Butalla, & Farinella, 2012; Maas & Farinella, 2012; Maas, et al., 2019), However,
  - These have had small sample sizes
  - There may be interactions of other factors, like age and severity

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Principles of Motor Learning*

Principle	Acquisition/Less Verbal Child	Retention/More Verbal Child
Practice Amount	More	Less
Practice Distribution	Mass	Distributed
Practice Variability	Consistent context, consistent prosody, pitch, rate	Varied context, varied prosody, pitch, rate
Practice Schedule	Blocked, predictable order	Random unpredictable order
Feedback Type	Knowledge of performance	Knowledge of results
Feedback Frequency	Often, immediate	Inconsistent, delayed
Rate	Slow	Normal, varied

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PML: Amount of practice

- Three CAS studies

(Edeal & Gildersleeve-Neumann, 2011; Maas et al., 2019; Namasivayam et al., 2015)

More trials facilitate greater retention

- More opportunities to figure out articulatory configurations, motor commands, movement outcome

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PML: Amount of Practice

Maximizing amount of practice:

Smaller target set, more trials per target

Fast reinforcers with fewer and shorter breaks

More sessions
- Enlist practice partners, where practical and possible

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PML: Practice Distribution

- Massed = minimal time between trials or sessions
- Distributed = greater amount of time between trials or sessions
  - Distribution ⍰ Intensity
- Equivocal evidence in non-speech literature.

  Distributed practice better for retention
- Three CAS studies (Maas et al., 2019; Namasivayam et al., 2015; Thomas et al., 2014)

  Massed practice facilitates acquisition

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PML: Maximizing Intensity (Massed Practice)

- More sessions per week, closer in time
- Fewer targets
- Identify priority target/targets for focus within a session

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PML: Variability

Constant = practice one consistent exemplar

Variable = modify rate, loudness, inflection, context

Me Me! Me? Me too!

Constant practice facilitates acquisition/performance, but variable facilitates learning (Lai, et al., 2000)

Constant followed by variable helps to establish pattern, then variable practice supports adapting to different contexts (Lai, et al., 2000)

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PML: Variability

One CAS study (Preston et al., 2017)

Variable practice resulted in improved outcomes

(but initial constant practice may be beneficial)

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PML: Managing Variability

- Vary phonetic context (e.g., single words vs sequence/phrases)
- Vary prosody (e.g., question/statement intonation)
- Vary other suprasegmentals (e.g., loud/soft, fast/slow)
- Vary elicitation tasks (e.g., repetition, picture naming)
- Vary physical location or communication partners

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PML: Practice Schedule

Blocked = all targets practiced together in predictable sequence

I do	I do
Mommy	Mommy
Bye	Bye
Up	Up

Random = order or presentation of targets varied randomly

I do	Mommy
Bye	Bye
Up	Mommy
I do	Up

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One CAS study (Maas & Farinella, 2012)

Blocked practice may be more beneficial for some children

- moving from blocked to random practice, especially in children with severe CAS, is consistent with Guadagnoli and Lee's (2004) challenge point framework

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PML: Feedback

Type: Knowledge of Results
- That was right! Those were all good!

Knowledge of Performance
- Start with your lips closed, Keep your mouth moving

Frequency
- Children may need high rate for best acquisition (Sullivan et al., 2002)

Delayed vs Immediate

Clinician controlled vs self-rating

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PML: Feedback

One CAS study (Maas et al., 2012)

- **low rate** of feedback advantage for 2 of 4 children
- The Rapid Syllable Transition (ReST) protocol uses low rate of feedback
- Sullivan et al. (2008) suggest that children may need more feedback and for longer than adults to acquire nonspeech motor skill

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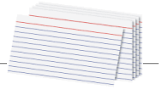
Managing Feedback



- Make flashcards depicting targets, written/pictured
- For High FB frequency:
  - Mark targets for KP or KR as needed for each target
  - Work towards KR for all
- For Low FB frequency:
  - For consistent, Low FB frequency: make 10 flashcards per target, mark 6 for FB. Shuffle cards.
  - For fading Low FB frequency: Create trial tracking sheet with some slots marked for FB

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PML in a Session

I want one X40
I do X1
Thomas X50
I do X1
Puppy X 40
I do X1
Time to go X5

Are these targets being practiced as…..

Massed or Distributed?

Constant or Variable?

Blocked or Random?

What type of feedback might you be giving?

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Remember Challenge Point

Child is producing a target easily. Options to increase challenge:

- Moving from mass to distributed practice
- Eliciting target with varied prosody
- Practicing randomly throughout session
- Changing from knowledge of performance to knowledge of results/reduce feedback
- Changing context (level of cueing, phonetic context/sequence, or physical conditions)

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Remember Challenge Point

Child is struggling to produce a target. Options to decease challenge:

- Returning to mass practice → accurate productions
- Maintain constant production
- Elicit target at predictable times in target list
- Give frequent feedback regarding performance
- Maintain context (level of cueing, phonetic context/sequence, or physical conditions)

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Principles of Motor Learning*

*Use Careful Clinical Judgment

(Manes & Robin, 2012)

	Acquisition/Less Verbal Child	Retention/More Verbal Child
Practice		Less
Practice Distribution		Distributed
Practice Variability		Varied context, varied prosody, pitch, rate
Practice Schedule	Blocked, pr… order	…unpredictable
Feedback Type	Knowledge of performance	…ults
Feedback Frequency	Often, immediate	In…
Rate	Slow	Normal, va…

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Treatment Approaches Strongest Evidence

- DTTC and other integral stimulation-based approaches (e.g., Edeal & Gildersleeve-Neumann, 2011; Maas, et al., 2012, 2014, 2019; Murray, McCabe, & Ballard, 2014; Strand, et al., 2006), Strand tutorial (2020)
- ReST (e.g., Ballard et al., 2010; Murray et al, 2014, 2015; Morgan et al., 2018; Mc Cabe et al., 2020)
- Ultrasound biofeedback treatment (e.g., Preston et al., 2013, 2016, 2017, 2020)
  - Speech Motor Chaining, Preston tutorial (2019)

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Treatment Approaches Strongest Evidence

- Integrated Phonological Awareness (McNeill, Gillon, & Dodd, 2009; Murray, McCabe, & Ballard, 2014)
- Motor Speech Treatment Protocol (e.g., Namasivayam et al., 2015)
- Nuffield (Morgan et al., 2018; Murray, McCabe, & Ballard, 2015)

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Additional Treatment Approaches

PROMPT (Dale & Hayden, 2013)

Melodic Intonation (Helfrich-Miller, 1994)

Kaufman (Gomez, 2018)

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Intervention Summary

- PML are the #1 ingredient in motor-based intervention for CAS
  - Always test for retention and transfer to measure motor ***learning*** (e.g., probe testing every third or fourth session)
- Keep Challenge Point in mind
  - Careful management of PML to maximize progress

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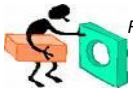
Take Home Message

Choose and modify intervention approach based on the child's presentation and progress



Fit the program to the child, not the child to the program

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A Challenge for You

Consider:

- learning to administer one new-to-you assessment test/task
- intentionally manipulating one or two PML factors to facilitate progress with a specific student/client (challenge point framework)
- learning more about an approach that you haven't used
- sharing something you learned with colleagues

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Related Issues: Comorbidities

- A notable prevalence of CAS was identified in 16p11.2, 22q11.2 deletion, Down syndrome, Galactosemia, and idiopathic intellectual disability (Shriberg et al., 2019)
  - "supports the efficiency of studying CAS in the context of complex neurodevelopmental disabilities."
- 16p.11.2 Raca et al., 2013
- 22q.11.2: Baylis & Shriberg, 2019
- Down syndrome: Rupela, Velleman & Adrianopoulos 2016; Wilson, Abbeduto, Camarata & Shriberg, 2019
- Galactosemia: Potter et al., 2013

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Related Issues: Comorbidities: ASD

- Shriberg et al., (2011): "no statistical support for hypothesis of concomitant CAS"
- Chenausky, et al. (2019): 13 of 54 low/minimally verbal participants categorized as suspected CAS.
  - May be a motor speech disorder similar to CAS but unique to minimally verbal children with ASD
  - May fit the Motor Speech Disorder-Not Otherwise Specified category of Shriberg et al. (2017).
  - May also reflect a lack of maturity (or practice)

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Related Issues: Multiple Languages

Assessment

Arabic: Abdou et al., 2020; Aziz et al., 2010

Cantonese: Wong et al, 2020

Treatment

Spanish: Gildersleeve-Neumann & Goldstein, 2015

- one of 2 children studied had CAS

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Related Issues: Motor Development

- Children with CAS performed below normal on all components of a movement assessment battery
- All children with CAS + LI were in the disordered category (compared to 1 of 3 children with CAS-only and 2 of 6 children with SSD + LI)

Suggests a higher order deficit that mediates cognitive-linguistic and motor impairments in CAS (Iuzzini-Seigel, 2019)

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Related Issues: Training Others to Treat

- Training parents for parent-clinician delivery resulted in variable treatment fidelity for ReST
- Less efficacious than previously reported clinician-only ReST (Thomas, McCabe, & Ballard, 2017)

- Training assistants in school to provide DTTC to two children showed positive results
- Assistants reported positive experiences (Lim, McCabe, & Purcell, 2019).

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Related Issues: Telehealth

"Telehealth delivery produced similar acquisition of pseudo-words and generalization to untreated behaviours as face-to-face delivery; however, following treatment, children showed stable rather than improving speech skills"

- "Caregivers and clinicians were satisfied with the telehealth treatment" (Thomas, McCabe, Ballard, & Lincoln, 2016)

Examination of parent perspectives (Thomas, McCabe, Ballard, & Bricker-Katz, 2018)

- Identified some positives and some cautions
- Clinicians need to consider multiple factors

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Related Issues: Literacy

- Phonological awareness is a key issue (Anthony, et al., 2011; Miller, et al., 2019; Tambyraja, Farquharson & Justice, 2020).
- Auditory Perception
  - On a measure of speech perception (syllable discrimination). Children with CAS + LI or SD + LI showed poorer discrimination, with appreciable within-group variability.
  - Support for the importance of accounting for language (Iuzzini-Seigel et al., 2015; Zuk, et al., 2018)

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Questions?

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Thanks to Dr. Adriana Lavi, conference organizer

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