THE CAS-ASD HYPOTHESIS
A CRITICAL EXAMINATION
OF THE EVIDENCE

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CAS-ASD HYPOTHESIS

Strong Form: CAS is a sufficient cause of lack of
speech development in at least some children
classified as nonverbal ASD

Weak Form: CAS contributes to the inappropriate
speech, prosody, and/or voice features reported in
some children and adults with verbal ASD
CAS DEFINITION

Childhood apraxia of speech (CAS) is a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone).

The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody.

ASHA 2007

3 FEATURES THAT HAVE GAINED SOME CONSENSUS AMONG INVESTIGATORS OF CAS

Inconsistent errors on consonants and vowels in repeated productions of syllables or words

Inappropriate prosody, especially in the realization of lexical or phrasal stress

Lengthened and disrupted coarticulatory transitions between sounds and syllables

STRAND 10-POINT CHECKLIST (SHRIBERG, POTTER, ET AL., 2009)

1. Difficulty achieving initial articulatory configurations and transitions into vowels
2. Syllable segregation
3. Lexical stress errors or equal stress
4. Vowel or consonant distortions including distorted substitutions
5. Groping (non-speech)
STRAND 10-POINT CHECKLIST
(SHRIBERG, POTTER, ET AL., 2009)

6. Intrusive schwa
7. Voicing errors
8. Slow rate
9. DDK rate
10. Increased difficulty with longer or more phonetically complex words

ASD – DEFINITION
DSM V

1. Deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation; to reduced sharing of interests, emotions, or affect; to failure to initiate or respond to social interactions.
2. Deficits in nonverbal communicative behaviors used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication.
3. Deficits in developing, maintaining, and understanding relationships, ranging, for example, from difficulties adjusting behavior to suit various social contexts; to difficulties in sharing imaginative play or in making friends; to absence of interest in peers.

ASD – DEFINITION
DSM V

Restricted, repetitive patterns of behavior, interests, or activities as manifested by at least two of the following:
1. Stereotyped or repetitive motor movements, use of objects, or speech
2. Insistence on sameness, inflexible adherence to routines, or ritualized patterns or verbal nonverbal behavior
3. Highly restricted, fixated interests that are abnormal in intensity or focus
4. Hyper- or hyposensitivity to sensory input or unusual interests in sensory aspects of the environment
ASD: ATYPICAL SPEECH AND VOICE CHARACTERISTICS

Articulation
Phonology
Rate
Repetitions and revisions
Pitch
Prosody
Loudness

WHAT COULD ACCOUNT FOR THE ATYPICAL SPEECH AND VOICE CHARACTERISTICS IN ASD?

Speech Attunement Framework

Social Feedback Loop

CAS

THE SPEECH ATTUNEMENT FRAMEWORK SHRIBERG, ET. AL. (2011)

The acquisition of articulate speech and appropriate prosody-voice requires a child to ‘tune in’ to the oral communications of the ambient community.

It also requires a child to ‘tune up’ the phonological and phonetic behaviors subserving intelligible and socially appropriate speech, prosody, and voice production.
SPEECH ATTUNEMENT FRAMEWORK SHRIBERG, ET. AL. (2011)

“Affective social reciprocity constraints might delay tuning in to the nuances in auditory events that must be imitated precisely . . .”

“Due to challenges in communication intent and social reciprocity, persons with ASD do not experience the pragmatic press to ‘tune up’ the precision of their speech, prosody, and/or voice.”

SPEECH ATTUNEMENT FRAMEWORK SHRIBERG, ET. AL. (2011)

“Some individuals with ASD have enhanced auditory perceptual ability underlying the propensity to ‘tune in’ to the acoustic features of speech . . . For the origin of speech errors, tuning in to speech early due to enhanced auditory perceptual propensity and capability might lead to speech production distortions, especially if there are some motor constraints that widen the gap between intention and capability (i.e., motor skill).”

SOCIAL FEEDBACK LOOP WARLAUMONT, ET. AL., (2014)

“Adult responses are more likely when child vocalizations are speech-related. In turn, a child vocalization is more likely to be speech related if the previous speech-related child vocalization received an immediate adult response. Taken together, these results are consistent with the idea that there is a social feedback loop between child and caregiver that promotes speech-language development.”
“When caregivers’ responses are contingent on infant vocalizations being speech-related (noncry, non-laugh, and non-vegetative) the result is more frequent speech-related child vocalizations.”

“Furthermore, when adults’ contingent responses are vocal, children’s future vocalizations acquire acoustic characteristics resembling the adults’, such as more speech-likeness, more vowel resonance, or better consonant-vowel timing.”

“Considering the huge number of child vocalizations that are produced within a day (on average well over 2,000) and accumulate over months and years, mutual contingencies between child and adult are expected to compound, contributing substantially to speech development.”

“ASD is associated with less social interaction, less frequent initiation of social interactions, and atypical turn taking patterns... (Children with ASD) also tend to produce fewer speech-related vocalizations, and their vocalizations tend to be atypical.”

ASD affects the feedback loop:

“First, children with ASD produce fewer vocalizations and/or fewer vocalizations that are speech-related. A reduction in vocalization rate leads to fewer iterations of the social feedback loop, reducing the number of child opportunities to learn from contingent social feedback.”

“Second, caregivers of children with ASD respond differently to their children’s vocalizations. Responses of adults interacting with children with autism are less contingent on the child vocalization being speech-related.”

“Third, the social feedback loop is diminished if children with autism have reduced ability to attend to and make use of contingent social responses.”

“Children with autism produce proportionally fewer speech-related vocalizations and the responses they receive are less contingent on whether their vocalizations are speech-related.”

“Such differences diminish the strength of the social feedback loop with cascading effects on speech development over time.”
CAS - ASD


VELLEMAN, ET. AL. (2010)

10 Children with ASD compared to children with suspected CAS and to typically developing children.

Age range of children with ASD: 4 – 6 ½ years old (with at least 50 word vocabulary)

Acoustic analyses revealed similarities and differences in the performance of children with ASD and those with suspected childhood apraxia of speech.

Limitation: The children in the ASD group were younger than those in the other groups. They also had a wide range of comprehension and attention difficulties.

SHRIBERG, PAUL, BLACK, VAN SATEN, (2011)

46 children aged 4 to 7 with Autism Spectrum Disorder (ASD) and intelligible speech compared to 3 study groups:

1. 10 Typically developing children (ages 4-7)
2. 13 children with speech delays (ages 4-6)
3. 15 children and adults with CAS
DATA
Digitally recorded speech samples were analyzed including phonetic transcription, acoustic analysis and prosody coding using the Prosody Voice Screening Profile, which looks at phrasing, rate, stress, loudness, pitch, voice quality, and resonance yielding 87 indices of precision and stability of speech, prosody and voice.

ANALYSIS
Acoustic analysis accomplished with software classified subtypes of speech impairment into the following categories:

SD (speech delay) (reduced intelligibility due to age-inappropriate deletions, substitutions and distortions)
SE (speech errors) (limited to sibilants and/or rhotics)
PSD (persistent speech sound disorder)
CAS (childhood apraxia of speech)
Dysarthria
MSD-NOS (motor-speech disorder not otherwise specified)

RESULTS
For the children with ASD:
SD: 15.2%
SE: 31.8%
CAS: 0%
MSD-NOS: 0%
PSD: 0%
DYSARTHRIA 0%
ASD SPEECH V. CAS

Participants with ASD did not have the significantly slow speech rate, lengthened vowels, and uncommon phoneme distortions that are core signs of motor speech disorders in adults and in contemporary research in CAS. Participants with ASD had voice differences not reported in CAS. The present study found that a statistically significant percentage of children with ASD had inappropriate loudness and inappropriate pitch. Differences in vocal pitch and loudness increasingly reported in autism have not been reported in descriptions of apraxia of speech.

DISCUSSION

The findings from this study do not support the hypothesis that CAS is a prevalent concomitant disorder in persons with verbal ASD. Moreover, they do not support the prevalence in the present sample of a less specific motor speech disorder termed Motor Speech Disorder-Not Otherwise Specified. Limitation: the study used continuous speech samples which might not have shown the motor deficits that become apparent in imitation tasks etc. The children with ASD all had intelligibility above 70%.

3 RATIONALES FOR CAS-ASD HYPOTHESIS

3 conceptual and empirical perspectives that motivate the CAS-ASD hypothesis:

1. Motor skills - persons with ASD have praxis deficits affecting a range of motor skills
2. Possibility of common genetic origins of CAS and ASD
3. Phenotypic similarity (speech characteristics are similar to CAS)
1ST RATIONALE

Motor skills – persons with ASD have praxis deficits affecting imitative processes and impairing acquisition of a range of motor skills.

Constraint: Speech is domain specific – the neural substrates of apraxia of speech differ from the neural substrates posited for other motor systems and other types of apraxia (e.g., oromotor apraxia, limb apraxia, ideomotor apraxia)

(Pickett et al. 2009) summarized findings from reports of 167 individuals with nonverbal ASD who acquired speech at age 5 or older. The speech findings in this discussion are not consistent with the signs of CAS.

2ND RATIONALE

Possibility of common genetic origins of CAS and ASD

Numerous candidate genes are of interest for ASD – only the FOXP2 gene has been associated with CAS

Another constraint on likelihood of gene comorbidity is the wide differences in their reported prevalence:

Ideopathic CAS: 0.1%-0.2%

ASD: 1%

Comorbid ASD and CAS expected to be extremely rare (i.e., 1/100,000, multiplying the individual probabilities of each disorder).

3RD RATIONALE

Speech, prosody, and voice characteristics of some children with low and high verbal ASD reportedly are similar to those found in children and adults with apraxia of speech.

Findings of present study do support this.

Prosody and voice differences between ASD and CAS
Deshmukh & Mccauley (2011)

12 children with high functioning autism (HFA)
11 children with motor-speech disorders (CAS)
11 children characterized as typically developing (TD)
Age range: 4 – 10 years old

Measures used

- Social Communication Questionnaire
- PPVT-4
- EVT-2
- OSMSE-3
- DEAP inconsistency subtest
- GFTA-2
- Conversation
- DDK task
- Multisyllabic word repetition (HAPP-3)
- Nonsense word repetition (NRT & SRT)

Skills assessed

- Oral motor and motor speech skills
- Articulation
- Accuracy, rate and consistency of target speech sound productions using stimuli and tasks targeting motor speech status
**DESHMUKH & MCCAULEY (2011)**

**Results**

The TD group performed the best.
The HFA group were intermediate.
The MSD group performed the poorest.
Only the differences between the MSD group and the other groups were found to be statistically significant.

The findings suggest that the HFA group were unimpaired in their motor-speech skills.

**DESHMUKH & MCCAULEY (2011)**

**Limitations:**

Only children with high-functioning autism participated in the study
Data was obtained only from structured tasks (no continuous speech sample)

**TIERNEY, ET. AL. (2015)**

Journal of Developmental Behavioral Pediatrics

The objective of this study was to determine the ability of The Checklist for Autism Spectrum Disorder (CASD) to accurately distinguish between children with apraxia and autism.

CASD is a 30-item checklist of autism symptoms, which can be completed within 15 minutes.
TIERNEY, ET. AL. (2015)

30 children referred to the Penn State Hershey Pediatric Developmental Communication Assessment Clinic for concerns regarding speech, language, or autism.

All 30 children had a developmental delay
All children were evaluated for ASD
A subset of the 30 children were evaluated by a speech language pathologist for apraxia (those children suspected by a developmental pediatrician as having apraxia).

TIERNEY, ET. AL. (2015)

Results:

11/30 children were diagnosed with ASD
19/30 children were diagnosed with CAS

7/11 (63.6%) of children diagnosed with autism also had apraxia

DISPARITY BETWEEN FINDINGS IN TIERNEY STUDY AND EXPECTED PREVALENCE OF CAS

ASHA CAS Practice Portal:

CAS is found in 3.4%-4.3% of the children referred for speech disorders (Delaney & Kent, 2004.)

63% (19/30) of the children in this study were diagnosed with CAS.
Disparity between findings in Tierney study and expected prevalence of ASD v. CAS

General population:
ASD: 1.0%
CAS: 0.1 – 0.2%
ASD is roughly 10 X more prevalent than CAS

This study:
ASD: 11/30
CAS: 19/30
CAS was 1.72 X (almost twice) as prevalent as ASD

Possible reason for these disparities

Participants’ age range was 24 to 55 months with a mean of 35.0

How many of the 19 children diagnosed with CAS in this study were under the age of 3?

How many of the 7 children diagnosed with CAS and Autism were under the age of 3?

Diagnosis under 3 years of age ASHA CAS Practice Portal

Diagnosis of CAS in children under 3 is challenging for a variety of reasons including:
The fact that some primary characteristics of CAS (e.g., word inconsistency, a predominant error pattern of omission, etc.) are characteristic of emerging speech in typically developing children under the age of 3 years;
The challenge of sorting out inability versus unwillingness to provide a speech sample or to attempt a speech target;
The possibility that changes occurring prior to age 3 (e.g., developmental maturation, social and linguistic peer exposure, and beneficial effects of therapy) may alter the diagnostic label.
ASHA CAS Practic Portal

Diagnosis below age 3 is best categorized under a provisional diagnostic classification: CAS cannot be ruled out, signs are consistent with CAS, or suspected to have CAS, rather than an unequivocal CAS.

TIERNEY, ET. AL. (2015)

Limitation: How the evaluation of CAS was conducted.
The CAS evaluation in this study consisted of a clinical interview with the parents and administration of the Kaufman Speech Praxis Test (KSPT) and the Preschool Language Scale-5 or Clinical Evaluation of Language Fundamentals. Apraxia diagnoses were based on performance on the KSPT and the presence of speech characteristics of apraxia observed by the speech and language pathologist during the language evaluation.


Reviewed the following:
- Apraxia Profile, Hickman 2000
- Kaufman Speech Praxis Test, Kaufman 1995
- Verbal Dyspraxia Profile, Jelm 2001
- Screening Test of Developmental Apraxia of Speech, Blakely 2001
- Verbal Motor Production Assessment for Children (VMPAC), Hayden & Square 1999

Kaufman Speech Praxis Test, Kaufman did not meet criteria for reliability or validity

“As part of their clinical decision making in children’s motor speech disorders, clinicians may continue to turn to standardized tests, but . . . Any of these methods requires cautious use and interpretation because of their unknown or rudimentary claims to reliability and validity.”

ASHA 2007

Lists necessary features of CAS evaluation. Some examples from the technical report include:
- differentiating children’s performance on functional/automatic versus volitional actions (counting 1-10 v. naming particular numbers; familiar phrases v. unfamiliar phrases)
- repetitions of the same stimulus versus repetitions of varying stimuli (e.g., sequential motion rates vs. alternating motion rates)
- tasks for which auditory versus visual versus tactile versus combinations of cues are provided

Assessment should include performance in multiple contexts (e.g., spontaneous, elicited, imitation; syllable, single-word, phrase, sentence, discourse)

“At present, no standardized test incorporates all of these features and those that have been formally critiqued have been found lacking in terms of important psychometric standards.”

AN EXAMPLE OF A MORE RIGOROUS ASSESSMENT PROTOCOL

Differential Diagnosis of Children with Suspected Childhood Apraxia of Speech
Elizabeth Murray, Patricia McCabe, Robert Heard, and Kirrie J. Ballard

Journal of Speech, Language, and Hearing Research
January 28, 2015.
AN EXAMPLE OF MORE EXACTING CRITERIA FOR DIFFERENTIAL DIAGNOSIS OF CAS  Murray, et. al., (2015)

Child must meet all of the following:
- Must present with all 3 of the core symptoms listed in the ASHA 2007 technical report in each of 3 speaking tasks
- Must present with at least 4 out of 10 features listed in the Strand 10 point check-list over 3 speaking tasks
- Inconsistency score of at least 40% on the DEAP Inconsistency subtest
- Test of Polysyllables:
  1. A minimum of 12 words with perceptually identified syllable segregations in the 50 word test.
  2. A minimum of 7 words with perceptually identified stress pattern errors.

CONCLUSION

Currently there is not sufficient or clear and convincing evidence that CAS contributes to the inappropriate speech, prosody, and/or voice features reported in some children and adults with verbal ASD

CLINICAL IMPLICATIONS OF THE CAS-ASD HYPOTHESIS

WHAT MUST THE PRIMARY THERAPY GOALS FOR YOUNG CHILDREN WITH ASD ADDRESS?
- Reciprocity
- Sustained engagement
- Shared attention
- Turn taking

COMMUNICATIVE INTENT
CLINICAL IMPLICATIONS OF THE CAS-ASD HYPOTHESIS

YOUNG CHILDREN WITH ASD

Therapeutic intervention must involve client-clinician interactions that focus on encouraging engagement, fostering reciprocity, and responding positively to communicative intent expressed by the child in a way that rewards it, and expands on it.

What must the primary therapy goal for young children with CAS address?
Improving motor-speech skills

Intervention Methods:
Motor-programing approaches
Integral stimulation (DTTC)
Tactile Kinesthetic approaches (Touch Cue, PROMPT)
Successive word approximation approaches

CLINICAL IMPLICATIONS OF THE CAS-ASD HYPOTHESIS

Young children with CAS
Therapeutic intervention must focus on client-clinician interactions that involve repetitive trial practice
Hundreds of trials per session
(Edeala and Gildersleeve-Neumann, 2011)
Imitative tasks with tactile cues and slow simultaneous models
For very young or severely impaired children: no more than 5 targets per session
Blocked practice in the beginning
(e.g. “pop” 20 x)
(Strand & Skinder, 1999)
ASD V. CAS
SOCIAL COMMUNICATION DEFICITS
V. MOTOR-SPEECH DEFICITS

Two very different sets of primary goals

Very different intervention approaches

The nature of the client-clinician interactions in the therapy room are very different

CLINICAL QUESTIONS

Is there sufficient or convincing evidence to support the CAS-ASD Hypothesis?

Should we be doing repetitive motor-speech drills with young children on the autistic spectrum who have significant deficits in social reciprocity, engagement, and communicative intent?

What would the relatively rigid, intense, motor-speech practice teach young children with ASD about social communication? What impact might it have on their social communication challenges?

Might we inadvertently exacerbate the primary deficits in social communication by ignoring acts of communicative intent that interfere or don’t coincide with the immediate motor-speech goals?

References


References


