



Examining the Reliability of Childhood Apraxia of Speech (CAS) Diagnostic Markers in the Greek Language: Preliminary Results

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ABSTRACT

Childhood apraxia of speech (CAS) is defined as “a neurological pediatric speech sound disorder in which the precision and consistency of movements underlying speech are impaired, in the absence of neuromuscular deficits. The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody.” [1]. Due to the absence of universal consensus on CAS diagnostic markers, it is significant to gain non-English clinicians’ insight on the subject. This study aims to present information regarding the reliability of three segmental and suprasegmental CAS specific speech characteristics that could be used to distinguish CAS from a Phonological disorder (PD) in the Greek language. Participants included 53 children ranging between 5;0 to 5;11 (years; months) with CAS (CAS, n=6), PD (n=7), CAS + PD (n=13), TD (typically developing, n=27). The Greek Phonetic and Phonological Development tool and specific subtests of the Verbal Motor production Assessment for Children (VMPAC) were administered, collecting data regarding sequencing, prosody and consistency of speech errors, during spontaneous and imitated production. Impaired prosody, speech error inconsistency and performance during sequencing tasks was significantly lower in participants with a CAS diagnosis. These findings demonstrate that the three

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features incorporated in ASHA's report [1] can indicate the direction for CAS differential diagnosis in the Greek language. It is suggested that the implementation of a broader set of diagnostic criteria would support a CAS diagnosis more efficiently, assisting in the differentiation of the above-mentioned disorders.

Keywords: childhood apraxia of speech, phonological disorder, differential diagnosis, speech characteristics.

INTRODUCTION

Speech sound disorders (SSDs) are among the most prevalent disorders in need of intervention in speech therapy clinics [2]. According to American Speech-Language-Hearing Association-ASHA, SSDs is a term used to cover a broad spectrum of pathologies, characterized by the individual's inability to produce appropriate speech sound sequences, past the expected age. This umbrella term refers to any singular difficulty or combination of difficulties, in motor production, perception or phonological representation of speech sounds and segments. Depending on the nature of the impairment, SSDs are categorized as organic, or functional (idiopathic). Organic SSDs refer to clinical entities, where their etiopathology lies in evident anatomical deficits, acquired or inherited. These include orofacial anomalies (e.g. trauma-induced, cleft/lip palate) or dysfunctional development of motor-neurological, such as childhood apraxia of speech (CAS) and sensory (e.g. hearing loss) pathways. Functional SSDs on the other hand, regard disrupted speech production of an inapparent cause, such as in the case of articulation and phonological disorders- PD [3].

Childhood apraxia of speech (CAS) is the specific SSD discussed in our study, the designated name used by ASHA and applied universally. According to ASHA:

CAS is a neurological childhood (pediatric) SSD 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 [1].

Various synonyms have been used historically, such as verbal dyspraxia, articulatory apraxia, developmental apraxia of speech, developmental verbal dyspraxia -a term used widely in the UK [4]- and CAS, with the latter adopted consistently worldwide [5-8]. Difficulties in diagnosing CAS, have led experts to use the term 'suspected CAS', instead of CAS [6]. In essence, individuals with this disorder experience difficulty in appropriately organizing the necessary muscles when speaking. This occurs, not because of some inherent weakness, but because of their brain's inability to direct and appropriately coordinate articulatory movements, which is imperative for comprehensive communication and normal socializing. The speech variables affected, are the ability to produce precise speech sounds and syllables, in the correct order and speech rhythm [9].

Although there has been substantial clinical and preclinical progress in the study of CAS, pathognomonic markers for CAS are yet to be identified. The limited sensitivity of current diagnostic tools, the overlapping symptoms of CAS with other disorders, the limited research base and the heterogeneity of the examinees (e.g., in terms of age, severity and co-existing

impairments) challenge the precise diagnosis of CAS. It should be noted that CAS occurs infrequently. It has been reported that CAS affects about 0,1% of the general population compared to other developmental speech disorders [10], such as articulation and phonological disorders which affect about 3.5% of preschool children [11]. These numerous challenges highlight the critical need for better CAS pathognomic markers.

Due to the absence of a consensus on the diagnostic markers for CAS, it is thus pivotal to gain experiences and scientific knowledge in diagnosis and assessment tools, from non- English clinicians as well, to determine whether CAS speech characteristics are seen universally [12]. Specifically, differences regarding phonetic and phonological characteristics of languages, may be associated with the underlying neurological deficit [13]. The above-mentioned study processed information derived from observations of infants that were later diagnosed with CAS. These children presented with fewer syllables per minute and fewer canonical babbles in comparison to other groups, and first achieved canonical babbling at a later age than expected. It is suggested that these results may be a pre-indication of their motor speech impairment.

If international consensus is achieved, accurate diagnosis and individualized intervention programs may be implemented [14]. While ASHA [1] noted that “there is no validated list of diagnostic features of CAS that differentiates this symptom complex from other types of childhood SSDs”, the CAS Technical Report proposed the following three segmental and suprasegmental speech characteristics to be established as CAS specific:

- 1 inconsistent errors of vowels and consonants during repeated productions of syllable or words
- 2 inappropriate prosody, especially when placing emphasis on a certain syllable or word
- 3 disrupted and lengthened co-articulatory transitions between sounds and syllables

In addition to the core features of CAS, children may present co-occurring impairments in non-speech oral motor function ([13], language [15], phonemic awareness and literacy skill development [16,17], in turn impacting negatively academic and occupational success [18]. CAS has been associated with various medical conditions, such as metabolic disorders [19], epilepsy [20], FOXP2 gene alteration [21], Koolen de Vries Syndrome [22] and others, whereas parameters possibly impacting treatment prognosis, are still under exploration.

It is essential to identify the relative contribution of motor planning and programming difficulties that a child with SSDs encounters, in order to achieve an accurate diagnosis. In the cases of children with severe SSD, speech-language pathologists perceive it as a major clinical challenge to discriminate, which errors are due to motor and which due to linguistic impairment [23], and the possible complex interactions as well. Additional difficulties emerging during the differential diagnosis of CAS, are presumably attributed to the fact, that the characteristics of this disorder appear to change with age and maturation [24].

Studies Aiming to Differentiate CAS from Other Speech Sound Disorders

Polat and Logacev [25] aimed to define speech characteristics that are CAS diagnostic in order to be incorporated in related tests and used in the distinction from PD. According to the findings, errors of intonation, vowel pronunciation and syllable stress are strong CAS indicators and inconsistent production and voicing mistakes can additionally facilitate differentiation. Murray et al [6] focused on determining a set of quantitative clinically derived measures, whose

combined evaluation could distinguish CAS from other speech disorders. Such an attempt is justified, considering that the current standard for CAS differential diagnosis relies on expert evaluation of a small number of speech behaviors and standardized clinical protocols are lacking. CAS diagnosis required 3 ASHA consensus-based features [1] and any 4 of the Strand's 10-point checklist [26], over at least 3 assessment tasks. Standardized tests examined oral motor control abilities and features like inconsistency, sound and syllable sequencing and lexical stress accuracy. Responses were used to form expert qualitative judgments on the existence or absence of CAS and generate 24 quantitative measures for statistical purposes. Findings indicated that accessing the combination of 4 measures strongly predicts CAS in verbal children, against the current gold standard of expert opinion.

A study by Grigos et al [27] explored the potential interrelation of CAS and oral articulatory control, by investigating aspects of speech motor control (temporal and spatial). Tasks with progressively increased linguistic complexity via increases in word length were applied. The findings support the proposition, that children with CAS can be distinguished from those with SSDs, based on movement variability, especially as task requirements increase. It also succeeds in illustrating, how challenging isolating parts of speech-motor processing really is and in recognizing speech-motor impairment, as an underlying factor responsible for inaccurate speech in CAS cases. On that note, it is suggested that motor and linguistic factor interrelationships should be further investigated, focusing on the modification of semantic, syntactic and phonologic complexity.

Bahr [28] tested whether examining articulatory gestures can assist in differentiating severe SSDs. The idea under investigation was that children with CAS may find transitioning between gestures more troublesome, than those with PD. It was confirmed that both CAS and PD groups experienced difficulty with gestures that involved the participation of the glottis and tongue blade, while performing in a similar manner in general. Interestingly, the CAS group demonstrated more flawed speech productions, when they involved lip movement combinations. They also appeared to display longer acoustic and kinematic duration, a possible indication of speech programming and coordination impairment and therefore requiring additional time to achieve articulatory targets [29].

The notion that individuals diagnosed with CAS present a higher degree of difficulty in facial muscle coordination during speech, when compared to children having other SSDs, was explored by Moss and Grigos [30]. Reflective markers were placed on the lips and jaws of the participants, enabling movement observation, during the pronunciation of three selected words (one monosyllabic and two multisyllabic). Based on the researcher's findings, individuals with CAS demonstrate more difficulty in achieving the needed coordination during demanding speech production compared to those of other groups. In another study Kopera and Grigos [31], investigated whether kinematic and acoustic parameters involved in producing the needed accented syllables for conveying appropriate meaning, differentiate CAS from other SSDs with overlapping symptoms, such as Speech Delay. Facial reflective markers were placed to monitor motor control during the examination. This research concluded that children with CAS displayed noticeable differences only with the ones with TD, as far as the length of the durational distinction between syllables emphasized and not, of what were perceived as accurate word productions, where the former groups specifically appeared smaller.

It is generally accepted that CAS is in essence, a motor speech disorder. Nevertheless, it has been supported that it could very well be a linguistic disorder of a broader deficit spectrum [32]. The high prevalence of co-existing language impairments amongst individuals with CAS supports this theory [33,34]. Experts have hypothesized, that these children produce phonetic segments of affected sequencing [35]. A study of interest was conducted by Iuzzini et al [36] with the purpose of tackling two lingering questions: 1- does speech inconsistency reflect a CAS core impairment or a co-existing Language Impairment (LI) and 2- can it be used as a reliable marker, to differentiate individuals presenting with CAS from those with SD. The findings confirmed that speech inconsistency is a core CAS characteristic, possibly resulting from deficits in articulatory planning and programming. This characteristic successfully enables distinguishing CAS from SD individuals [37]. In cases bearing an LI, the phenomenon could be attributed to a higher-level language disability [35].

According to Maas [9], the complexity of articulatory movement across combinations of phonemes within syllable sequences, has been an unclearly defined variable. Case and Grigos [38] acknowledge the need to determine the underlying structure of motoric interrelationships. It views complexity, based on movement demands and how it affects speech performance and not at the level of isolated phonemes as seen in preceding research. What had been hypothesized, was that levels of motoric complexity may require measures of speech motor control to be determined and that accuracy becomes challenging with multi-syllable sequences in individuals with CAS. Decreased consonant vowel accuracy, an extended movement duration and a greater level of kinematic and acoustic variability, was presumed to reflect increased motor complexity. All children's performance was impacted when movement demands increased, as reflected by longer movement duration and greater motor variability. It also provided strong evidence of the importance of taking under consideration motoric elements, when conducting speech production evaluations and designing relative testing stimuli.

Difficulty is apparent in differentiating CAS from other SSDs, predominately due to phonological-level delay, due to the fact that children tend to make similar verbal mistakes. Aziz et al [39] explored if individuals with CAS exhibit notable differences in certain skill performances, when compared to others with Multiple Phonological Disorders- MPD and to those with no communication disorder whatsoever. Based on the study's findings, identification of typical and non-typical CAS features was enabled and insight into interaction between language and speech performances, was obtained. What was considered essential in CAS differential diagnosis was the inclusion of variables, such as impaired prosody, ability to process syllabic data, as well as troublesome consonant cluster pronunciation. Evaluation of numerous cases, utilizing spontaneous and single word productions should be included.

Keske – Soares et al [40], investigated the differentiation of CAS from PD with the use of the Brazilian adaptation of the DEMSS test. Both disorders may share symptoms, such as impaired utterance of consonants and slow development, making differential diagnosis difficult to achieve. Some characteristic features of CAS are considered to be articulatory groping and multiple mistakes in pronouncing vowels. Nevertheless, a fundamental variable that outstands in CAS seems to be the fact that articulation deficits tend to linger on and subside in later stages of the child's life, in comparison to other speech disorders. The study concluded, that children with CAS present speech consistency and accuracy of a smaller degree, compared to those with PD and TD.

The need of identifying possible CAS presence in Arabic speakers was addressed by Abdou et al [41]. According to the research's findings, participants with CAS displayed high scoring in receptive language and low in expressive, as well as impaired prosody and a slower diadochokinesis (DDK) rate comparing to PD. Inconsistent mistakes were evident when producing the same utterances repeatedly, whereas indications of oral apraxia were absent when performing oral non-speech motor tasks. Ziethe et al [42] investigated whether the detection of mistakes in pronouncing vowels and consonants of various types and number, enables the differentiation of individuals with CAS from those with PD, utilizing acoustic and perceptual analysis. It was determined that there are significant differences regarding vowel and consonant articulation, between said groups and should be considered in differential diagnosis, alluding to the existence of particular speech motor planning and realizing impairment.

Overby et al [43,44], conducted two interesting studies, where home video footage of infants was examined in retrospect, aiming to recognize possible deviations of vocal development, of infants and toddlers, later diagnosed with CAS and SSD. The inability to vocally explore in the early stages of life (infants producing sounds of lesser variety and frequency than expected) has been associated with impaired motor speech control in children with CAS [45]. Considering the lack of existing studies with similar objective - target groups and the proven importance of examining early vocalization in defining the developmental characteristics of a communication disorder, this approach could assist in recognizing early manifestations of CAS. It specifically aimed at determining possible "red flags" in identifying the disorder. In both studies for the purpose of facilitating diagnosis of CAS, a version of the Mayo Clinic System's pediatric adaptation was implemented [26]. According to the above, the individual that exhibited at least 4 signs of troublesome speech motor planning and programming over 3 or more speech tasks, would be diagnosed with CAS.

The first study focused on comparing consonant emergence, volubility and syllabic structure. Although the study had certain limitations and the results were at a preliminary stage, it was evident that children with CAS and SSD displayed different speech sound development than the TD group. Specifically, they presented lower volubility and diverse phonemic range. A common possible red flag seemed to be the slower appearance of the first resonant consonant. The second study's purpose was to recognize possible deviations in two measures of vocal development, referring to volubility and canonical babbling. Infants that were later diagnosed with CAS presented fewer syllables per minute and canonical babbles in comparison to other participants, and first achieved canonical babbling at a later age than expected. It is suggested that these results may be a pre-indication of their motor speech impairment.

Purpose of Current Study

The primary aim of this study is to determine the features that would decisively assist in distinguishing CAS from other SSDs, specifically from a severe phonological disorder, in the Greek language. It should be noted that available Greek standardized assessment tools for CAS are extremely limited. Also, our goal is to present information that could be useful in the effort of establishing an international consensus in diagnostic markers for CAS. Existing research investigating the features generally accepted as core CAS symptoms and therefore the differentiation from other SSD is limited by small sample sizes. Based on the premise that CAS rarely occurs alone and is often accompanied by other types of speech and language deficits, it

is essential for future studies to include a comparison group of children with comorbid suspected CAS and other speech sound impairments. Moreover, in order to achieve an accurate, diagnose of CAS, we utilized a combinatorial evaluation approach, consisting of the interpretation of the observations made by clinicians with knowledge and experience in the CAS area (subjective assessment), as well as the implementation of valid and reliable protocols (objective assessment) [40]. During the investigation, the information derived from the administration of standardized tools, was combined with possible correlations of speech behaviors associated with suspected CAS diagnosis, evident to the clinicians.

METHODS

Design and Participants

This study is part of a research project conducted at the National and Kapodistrian University of Athens' School of Medicine and ethical approval was obtained by the Ethical Review Board of the Eginition Hospital. Prior to the implementation of the assessment tools, specific authorization of the participants was requested by the legal guardians, who signed an Informed Consent Form, receiving explicit information concerning the purpose and implementation of the study and how collected data would be stored. The routine assessment protocol included patient interview and history taking, to obtain information concerning feeding difficulties, age of babbling and word emergence, as well as prenatal, natal or postnatal relevant medical problems. All the participants were assessed individually in public and private pediatric speech therapy clinics and preschools from 2018 to 2020.

Fifty-three children (N=53, M age= 5.46 years old, SD= 0.26 years old, range 5 to 6 years old) were recruited for this study, twenty-four males and twenty-nine females. These participants were drawn from a larger investigation concerning the differentiation criteria of CAS and other SSDs and were age-matched. All of the children completed a standardized cognitive and language testing protocol, as well as a hearing screening, as part of their participation in the larger study. Exclusionary criteria were cognitive delay, structural deficits, evidence of dysarthria, syndromes and hearing or visual impairment. Inclusionary criteria were severe difficulties during speech sound production, the degree of which varied within the groups. Participants were categorized in 4 possible subject-groups: Group 1: CAS, N=6; Group 2: phonological disorder PD, N=7; Group 3: CAS and comorbid phonological disorder, N=13; and Group 4: control group without any communication disorders, N=27. All participants were monolingual Greek speakers. Age, gender and socioeconomic distribution were matched between the 4 groups.

CAS group (Group 1): The diagnostic classification for the CAS group was determined according to the presence of the three core features of the disorder, in more than one speaking context, as highlighted in the ASHA [1] position statement: inconsistent errors across repeated productions, disrupted and lengthened coarticulatory transitions between sounds and syllables, as well as inappropriate prosody. Inconsistent errors were defined as consonant and vowel errors that differed across repeated productions of the same word. Difficulties forming articulatory transitions were characterized by poor sequencing of sounds or syllables, particularly when they included phonemes that were present in the child's repertoire, whereas prosodic errors were identified as incorrect lexical and/or phrasal stress. In addition to the three core features, children with suspected CAS demonstrated the following characteristics:

timing errors, phoneme distortions, inadequate diadochokinetic profile, articulatory groping and impaired volitional oral movement.

PD group (Group2): The assignment to the PD group was determined with the demonstration of consistent errors (consonant/vowel substitutions, omissions, additions), as well as the systematic use of phonological processes consistent with a phonological impairment. Respectively participants in this group presented the aforementioned error patterns in more than one speaking context and did not exhibit a combination of the three features highlighted in ASHA's [1] position statement.

CAS and comorbid PD group (Group 3): Participants were assigned to the CAS and comorbid PD group, when displaying simultaneously prementioned features that bore CAS and PD diagnosis.

Control group (Group 4): The control group consisted of typically developed children that did not display any cognitive, language or speech impairment according to their test results and were chosen from private preschools.

Procedure and Measures

Based on the premise that the gold standard for CAS diagnosis is expert clinical opinion [9,6], the participants were classified into the above-mentioned groups according to clinical judgment of speech-language therapists, with prior experience in CAS assessment. The Phonetic and Phonological Development standardized test (Greek version) [46], auditory perception and the three consensus-based ASHA CAS features were utilized. Subsequently each participant was requested to complete speech and non- speech oral motor tasks, across three thirty-minute sessions, to avoid possible response degradation due to fatigue occurring from prolonged sessions. The responses during speech tasks were audio recorded, to reevaluate the scorings, when deemed necessary and for phonetic transcription. During the speech tasks, the participants were requested to produce test words- utterances spontaneously and imitatively. The tasks administered were components of a standardized assessment tool, the Verbal Motor production Assessment for Children- VMPAC [47].

VMPAC: The VMPAC is an assessment tool for children with speech sound disorders, which is stated to be given as part of a total assessment battery and has reliable normative data for English speaking children. The implementation of the test aims to provide information regarding the neuromotor integrity of the speech production system in children aged 3 to 12 years old who have speech sound disorders, as well as determining whether a speech motor issue underlies a child's speech sound disorder. Specifically, the information derived concerns the speech production system at rest and when engaged in vegetative and volitional non-speech and speech tasks. The items involve tasks evaluating Global motor and Focal oromotor control, Sequencing, Connected speech and Language control and Speech characteristics. Primarily all of the subtests of the VMPAC were implemented, in order to determine possible correlations concerning the parameters under evaluation.

The Global motor control section includes 20 items that address basic processes which underlie postural, respiratory and phonatory support, cerebral inhibition of oral reflexes and the development of coordinated vegetative functions. The Focal oromotor control area consists of

46 speech and non- speech tasks, in order to evaluate the child's volitional oromotor control of three speech subsystems, in isolation and in combination: mandibular, labial-facial and lingual. The area of Sequencing, is designed to assess the participant's ability to produce speech and non-speech movements in the correct sequential order and is comprised of 23 items. The Connected speech and Language control section, contains 5 items and assesses possible variability of the child's speech production, due to the complexity of language formulation, taking into consideration the precision and coordination of jaw, lip and tongue movements and their interactions (co-articulation). Finally, the Speech characteristics area of VMPAC, focuses on the evaluation of the child's pitch, resonance, voice quality, intensity, prosody, rate, as well as the speech production during automatic verbal sentences.

Greek Phonetic and Phonological Development test: The Greek Phonetic and Phonological Development tool is a standardized and reliable picture naming test designed for Greek speaking children aged 2,6-12 years old. A set of 101 target words, is included, with increasing syllable structure, of which 70 are elicited with the use of corresponding pictures, whereas 31 words are elicited with the use of 2 multisensory pictures. The implementation of the specific tool, enables the clinician to obtain information referring to the child's phonetic repertoire and speech sound production in initial, middle, and final word positions, as well as the analysis of the phonological system and possible phonological deviations, discrepant with the chronological age. Also, it provides a direction concerning speech error inconsistency.

The selection of specific subtests of the VMPAC was based upon the three core features of the CAS disorder, highlighted in the ASHA [1] position statement. Specifically, inconsistency in speech errors, was investigated with the administration of the Single (S.O.P.S.M) and More Single Oromotor- Phoneme Speech Movements (M.S.O.P.S.M) tasks of the Focal oromotor subtest. Difficulties with coarticulatory transitions were examined with the implementation of sequencing tasks, characterized with different levels of complexity, consisting of the Double (D.O.P.M), Triple (T.O.P.M), More Double (M.D.O.P.M), and More Triple Oromotor- Phoneme Speech Movements (M.T.O.P.M), tasks, the Oromotor Production in Word Sequences and Sentences (O.P.W.S.S), as well as the Oromotor Production in Automatic Verbal Sequences (O.P.A.V.S) task. Prosodic disruptions were explored with the use of the Speech characteristics subtest of the VMPAC.

Statistical Analysis

Quantitative variables were expressed as mean (+/- Standard Deviation) or as median (interquartile range). Qualitative variables were expressed as absolute and relative frequencies. For the comparison of proportions Fisher's exact tests were used. Analysis of variance (ANOVA) or non-parametric Kruskal-Wallis and Mann-Whitney tests were used for the comparison of continuous variables among the three groups. Post-hoc comparisons with Bonferroni correction were used in case of multiple testing in order to control for type I error. All reported p values are two-tailed. Statistical significance was set at $p < 0.05$ and analyses were conducted using SPSS statistical software (version 26.0).

RESULTS

The present study examined sequencing, prosody and consistency of speech errors, during spontaneous and imitated production, parameters when impaired, considered to indicate a speech motor issue underlying a child's speech sound disorder according to ASHA'S position

[1]. The features were tested multiple times within different manipulations. A descriptive test, the Greek Phonetic and Phonological Development test, evaluating phoneme error inconsistency was used, aiming to the classification of the participant groups of interest, CAS, PD, CAS+ PD and typical phonological development (TD) (Table 1).

Table 1: Consistency of errors, by group, according to the Greek Phonetic and Phonological Development test

No mistakes (TD)	27 (100.0)	0 (0.0)	<0.001+
Inconsistent errors (CAS)	0 (0.0)	6 (23.1)	
Consistent errors (PD)	0 (0.0)	7 (26.9)	
Inconsistent + consistent errors (CAS+PD)	0 (0.0)	13 (50)	

For the purpose of the article, the focus is restricted primarily to the participants depicting features of CAS and PD individually, as well as simultaneously. Sample consisted by 26 children (14 males and 12 females) with mean age 64.7 months (SD=4.65 months). Six children (23.1%) were in CAS group, seven (26.9%) were in PD group and 13 (50.0%) in the CAS+ PD group. Similar age and gender had the three groups of the participants (Table 2).

Sequencing ability was examined with the implementation of the Double (D.O.P.M), Triple (T.O.P.M), More Double (M.D.O.P.M), and More Triple Oromotor- Phoneme (Speech) Movements (M.T.O.P.M), tasks, the Oromotor Production in Word Sequences and Sentences (O.P.W.S.S), as well as the Oromotor Production in Automatic Verbal Sequences (O.P.A.V.S) task of the VMPAC test, characterized with different levels of complexity. Consistency of speech errors, during spontaneous and imitated production, was examined with the administration of the Single (S.O.P.S.M) and More Single Oromotor- Phoneme Speech Movements (M.S.O.P.S.M) subtasks, whereas prosody was assessed with the respective VMPAC task (Table 3).

Our statistical analyses show that T.O.P.M, (M)D.O.P.M, O.P.W.S.S, O.P.A.V.S., PROSODY and M.S.O.P.S.M differed significantly among the three groups (Table 3).

Table 2: Gender and age, by group

	Groups			P
	CAS (N=6; 23.1%)	P.D. (N=7; 26.9%)	CAS+ P.D. (N=13; 50%)	
	N (%)	N (%)	N (%)	
Gender				
Males	4 (66,7)	3 (42,9)	7 (53,8)	0.876++
Females	2 (33,3)	4 (57,1)	6 (46,2)	
Age (months), mean (SD)	67,8 (2,3)	65,6 (4,1)	62,8 (5)	0.068+

+ANOVA; ++Fisher's exact test

Table 3: Sequencing, Prosody and Error Consistency by group

	Groups						P
	CAS		PD		CAS+ PD		
	Mean	Median	Mean	Median	Mean	Median	
	(SD)	(IQR)	(SD)	(IQR)	(SD)	(IQR)	
SEQUENCING							
D.O.P.M(6)	4.7 (0.5)	5 (4 – 5)	5.3 (1)	6 (4 – 6)	4.5 (1.3)	5 (3 – 6)	0.295+
T.O.P.M(8)	2.8 (0.8)	3 (2 – 3)	6.4 (1.7)	7 (6 – 8)	3.8 (2.5)	4 (2 – 6)	0.015+

(M)D.O.P.M (6)	2.7 (1.2)	2.5 (2 – 4)	5.3 (1)	6 (4 – 6)	3 (2.3)	2 (1 – 6)	0.034+
(M)T.O.P.M (4)	0.7 (0.8)	0.5 (0 – 1)	1.1 (1.1)	1 (0 – 2)	0.9 (1.3)	0 (0 – 2)	0.689+
O.P.W.S.S (12)	3.2 (1)	3.5 (2 – 4)	9.9 (2)	10 (8 – 12)	5.8 (2.6)	5 (5 – 7)	0.001+
O.P.A.V.S. (1)	0 (0)	0 (0 – 0)	1 (0)	1 (1 – 1)	0.8 (0.4)	1 (1 – 1)	0.001+
PROSODY, N (%)							
0	6 (100)		0 (0)		10 (76.9)		<0.001++
1	0 (0)		7 (100)		3 (23.1)		
CONSISTENCY							
S.O.P.S.M (5)	4.3 (0.8)	4.5 (4 – 5)	5 (0)	5 (5 – 5)	4.3 (1)	5 (4 – 5)	0.133+
M.S.O.P.S.M (6)	4 (0.9)	4 (3 – 5)	6 (0)	6 (6 – 6)	5 (1.4)	6 (5 – 6)	0.004+

+Kruskal-Wallis test; ++Fisher's exact test

In particular, we observed that while D.O.P.M results allowed us to detect significant differences between all CAS subjects (CAS and CAS+PD groups) and the TD group, these measurements were not able to isolate the CAS group from the PD group (Figure 1).

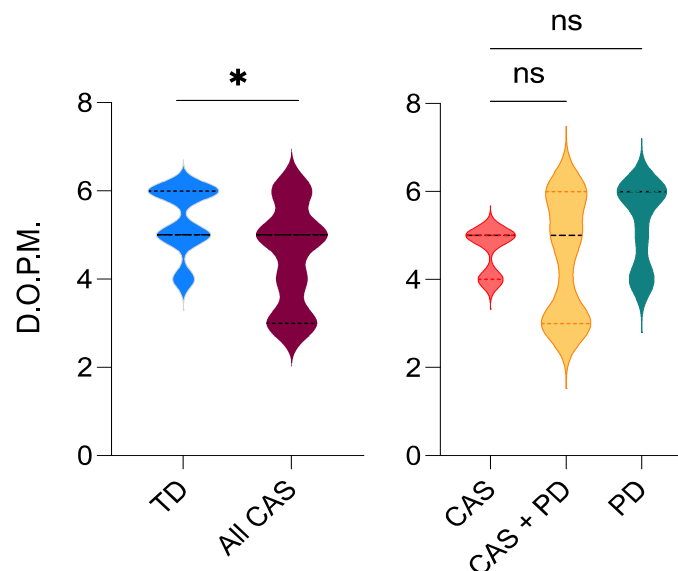


Figure 1: D.O.P.M. by group

Note. DOPM measurements show that the TD group has a higher score when compared to all individuals with CAS (i.e., CAS and CAS+PD) (Mann-Whitney, $p=0.012$, $N=27, 19$). Yet, D.O.P.M. assessments don't show statistical significance between the CAS and the PD groups (Kruskal-Wallis, $p=0.295$, $N=6-13$). Data are shown as violin plots.

As mentioned above, the assessment of T.O.P.M not only allowed us to observe significant differences between the TD groups and all the subjects with CAS ($p=0.0001$), but also allowed us to isolate the CAS group from children with PD. Indeed, after correction, for multiple comparisons, it was found that the PD group had greater T.O.P.M ($p=0.007$) compared to CAS (Figure 2).

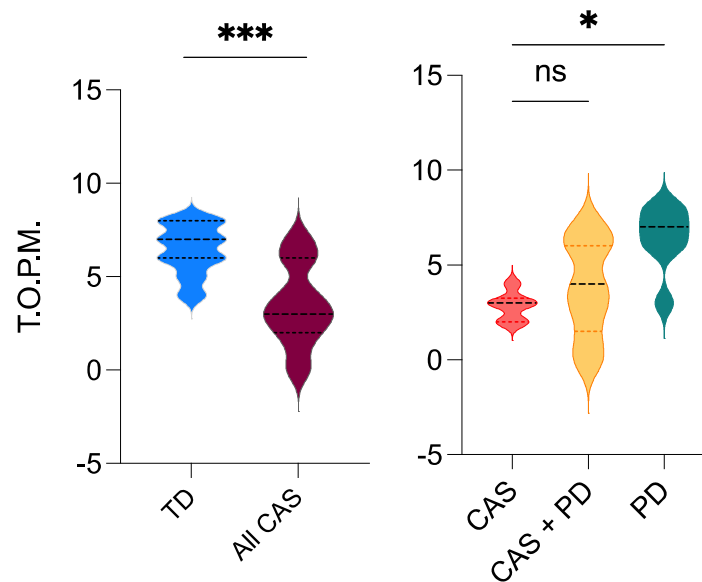


Figure 2: T.O.P.M. by group

Note. TOPM measurements show that the TD group has a higher score when compared to all individuals with CAS (i.e., CAS and CAS+PD) (Mann-Whitney, $p < 0.0001$, $N = 27, 19$) and show statistical significance between the CAS and the PD groups (Kruskal-Wallis, $p = 0.015$, $N = 6-13$).

Moreover, we observed significant differences in (M)D.O.P.M. among the groups tested (3). In particular, it was found that the PD group had significantly greater (M)D.O.P.M. ($p = 0.045$) compared to the CAS group (Figure 3).

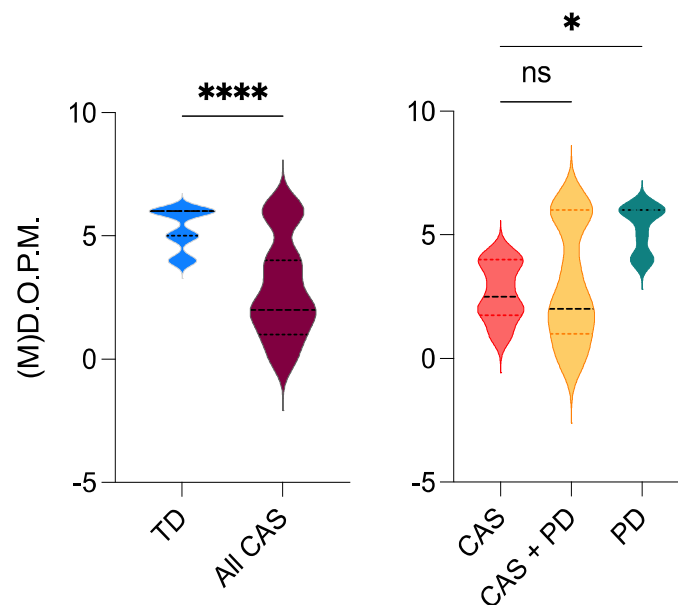


Figure 3: (M)D.O.P.M. by group

Note. TD group has a higher score when compared to all individuals with CAS (i.e., CAS and CAS+PD) (Mann-Whitney, $p < 0.0001$, $N = 27, 19$). Analysis shows statistical significance between the CAS and the PD groups (Kruskal-Wallis, $p = 0.045$, $N = 6-13$).

Additionally, we observed differences between the groups tested following the O.P.W.S.S task. The PD group had significantly greater O.P.W.S.S compared to CAS ($p=0.002$) and to CAS+ P.D. groups ($p=0.005$). Moreover, the CAS+ P.D. group had significantly greater O.P.W.S.S compared to the CAS group ($p=0.017$) (Figure 4).

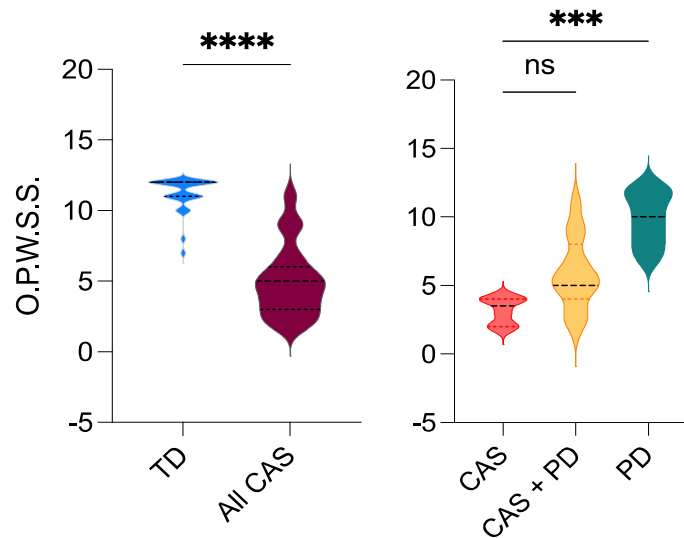


Figure 4: O.P.W.S.S. by group

Note. TD group has a higher score when compared to all individuals with CAS (i.e., CAS and CAS+PD) (Mann-Whitney, $p<0.0001$, $N=27, 19$). Analysis shows statistical significance between The CAS and the PD groups (Kruskal-Wallis, $p=0.045$, $N=6-13$).

We also observed that O.P.A.V.S. was significantly greater in the PD group compared to CAS ($p=0.001$). Yet our analyses also show significant differences between CAS and CAS+ P.D. groups ($p=0.002$). Together these results suggest that O.P.A.V.S assessment may not be sufficient to fully isolate individuals with CAS, when CAS co-occurs with other SSDs (Figure 5).

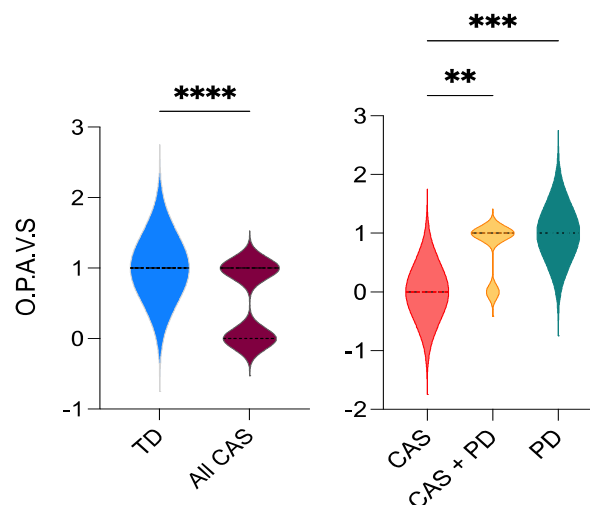


Figure 5: O.P.A.V.S. by group

Note. TD group has a higher score when compared to all individuals with CAS (i.e., CAS and CAS+PD) (Mann-Whitney, $p < 0.0001$, $N = 27, 19$). The analysis also shows statistical significance between the CAS and the CAS+PD and PD groups ($p = 0.001$ and $p = 0.002$, $N = 6-13$).

Furthermore, a similar pattern to O.P.A.V.S. results was observed during our M.S.O.P.S.M assessment (6). In particular, we observed that PD individuals display a significantly higher O.P.A.V.S. score than CAS subjects ($p = 0.001$). Yet the CAS+PD group was not significantly different than the PD group (Figure 6).

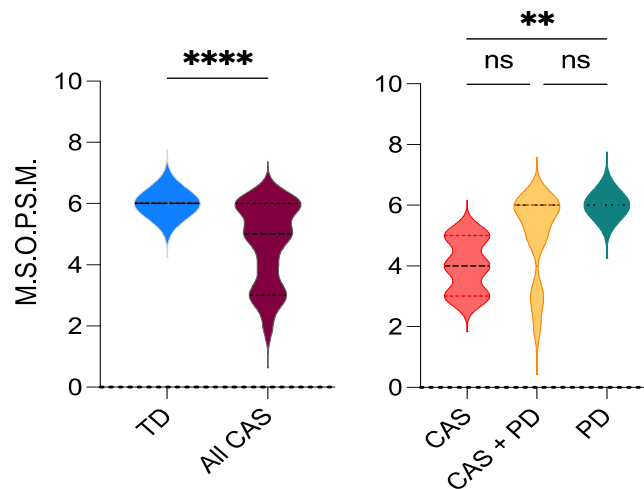


Figure 6: M.S.O.P.S.M. by group

Note. TD group has a higher score when compared to all individuals with CAS (i.e., CAS and CAS+PD) (Mann-Whitney, $p < 0.0001$, $N = 27, 19$). The analysis also shows statistical significance between the CAS and the PD groups ($p = 0.001$, $N = 6-13$).

Lastly, the percentage of PROSODY was significantly higher in the PD group compared to CAS and CAS+ P.D. groups ($p = 0.001$ and $p = 0.003$ respectively)(Figure 7).

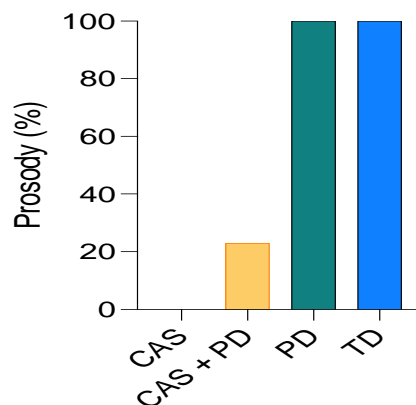


Figure 7: Prosody by group

Note. The TD group has a higher score when compared to all individuals with CAS and CAS+PD. We can't observe significant differences between the PD and the TD groups.

As observed in Table 4, CAS and CAS+PD groups exhibited more difficulties with coarticulatory transitions, during the majority of the sequencing tasks, compared to the PD and TD groups, which presented similar higher percentages. However, significant deviance was not portrayed between the aforementioned groups during the Double- Oromotor Phoneme Movement task (D.O.P.M), as well as the More Triple Oromotor- Phoneme Movement (M)T.O.P.M task.

Table 4: Percentages of performance in sequencing, prosody and error consistency tasks, by group

	Sequencing				Prosody		Error consistency		
	D.O.P. M	T.O.P. M	(M)D.O.P. M	(M)T.O.P. M	O.P.W.S .S	O.P.A.V.S .	S.O.P.S. M	M.S.O.P.S. M	
CAS	77.60	35	44.56	16.50	27.66	0	0	88.60	66.67
P.D.	88.17	80.36	88.17	28.50	82.17	100	100	100	100
CAS	74.33	47.13	50	23	48.75	77	23	86	83.33
+ P.									
D									
T. D	88.30	80.50	88.33	29.75	92.92	100	100	100	100

In general, sequencing skills of the CAS group, followed by the CAS+PD group, were characterized by poor performance, compared to the PD and TD groups, which presented similar findings, with the exception of the Oromotor production in the Automatic Verbal Sequences task (O.P.A.V.S.), according to which the CAS group acquired a zero score. Isolated vowel and consonant imitated production skills were tested by targeting the sounds /a, i, u, o, m, p, b, t, d[^], k, g[^]/.

Accordingly, all the participants in the PD and TD groups articulated the target phonemes correctly. On the contrary, the participants in the CAS and CAS+PD groups exhibited lower performance, presenting a more significant difficulty articulating the target sounds / p, b, t, d[^], k, g[^]/.

DISCUSSION

The purpose of the present investigation was to examine reliable markers that would facilitate diagnosis of CAS in the Greek language and therefore assist in the distinguishment of CAS from a severe phonological disorder. For this purpose, specific subtests of the VMPAC were administered. This allowed us to assess speech characteristics associated with CAS diagnosis, reported in other studies, as well as in the ASHA [1] position statement. The speech features include difficulty with co-articulatory transitions, prosodic disturbance, and inconsistency of speech sounds. Previous research findings suggest that CAS rarely occurs on its own [48-52] and there may be an overlap of speech difficulties between disorders [53]. Additionally, there is a significant correlation concerning motor and cognitive-linguistic systems during development [54]. Based on this premise a comparison group of children with comorbid CAS and PD, was included, in addition to the CAS, PD and TD groups, which were respectively matched in terms of chronological age, in order to make a valid comparison.

We observed that the lowest scores during sequencing tasks were exhibited by participants in the CAS group, followed consecutively by the CAS+ PD, the PD and the TD groups. These results are consistent with the literature and show that disrupted transitions between sounds and

syllables may result of impaired planning/programming of speech movements. Such measurements facilitate drawing diagnostic conclusions about the presence or absence of CAS [1]. The administration of the D.O.P.M, T.O.P.M, M.D.O.P.M and M.T.O.P.M aim to assess the participants' ability to accurately sequence multiple double and triple vowel-vowel and consonant-vowel syllables in novel tasks that do not rely on pre-established motor plans, without requiring prosodic accuracy. It was predicted and confirmed that children with CAS would encounter more difficulty in imitating speech productions compared to the PD group. However, a striking finding is that the CAS performance during the implementation of the D.O.P.M was better than the CAS+PD. The D.O.P.M is the only one in which the CAS group performed better than the CAS+PD group throughout the assessment. Additionally, significant deviance was not portrayed between the performances of the CAS group and the other groups during the (M.) T.O.P.M task, where the imitation of trisyllable consonant- vowel non-words was requested.

According to the literature, increasing utterance length, results in increasing difficulty which can be considered an indicator of CAS [25,29,55,56]. Based on this hypothesis, the initial sequencing tasks consisted of the imitation of isolated word productions that increased in length, providing information of possible difficulties during the transitioning between sounds and syllables. Due to the fact that the spatiotemporal planning/programming of speech gestures in connected speech is considered to be a more complex task than the planning/programming of gestures in isolated words [57], the O.P.W.S.S task that was sequentially administered, evaluated the oromotor production of word sequences and sentences, during which the participants were asked to imitate directly specific utterances, with the use of pictures. The CAS group presented a significantly lower performance compared to the other pathological groups. Additionally, as predicted CAS participants exhibited lower scores when syllables in speech stimuli increased, during the imitation of non-words, used in the D.O.P.M, T.O.P.M, M.D.O.P.M and M.T.O.P.M tasks. Also, it should be noted that they encountered greater difficulty imitating disyllable and trisyllable consonant-vowel novel words, compared to vowel-vowel words. This was noticeable as well for the CAS+PD group; however, they exhibited a better performance compared to the CAS group. In the section of O.P.A.V.S., the PD and TD groups presented the highest scores, indicating that their motor control was the same in automatic speech and self-formulated speech. Contrastingly, the CAS participants' speech motor movements were better in automatic sequences, compared to unfamiliar utterances and, therefore acquiring a zero score, succeeded by the CAS+PD group with a significantly higher score. Although there is increasing recognition that CAS and PD do co-occur [58], future investigations ought to further investigate in what ways language and phonological processing ability influence sound sequencing performance in the Greek language, taking into consideration that speech production is a multidimensional activity, featuring motoric and linguistic aspects.

One of the main features, considered to indicate a speech motor issue underlying a child's speech sound disorder and accepted by ASHA and other researchers, is inappropriate prosody [39,1]. The findings of this study indicate that prosody is significantly affected when bearing a CAS diagnosis, a premise which is underpinned by the low performances of the CAS and CAS+PD groups.

Prior studies confirm that speech inconsistency is a core CAS characteristic [37]. It is considered that inconsistency can be associated with difficulty consistently planning and programming the appropriate direction, force, timing, and gradation of articulatory movements and therefore consists of a predominant feature used to differentiate CAS from nonmotor-based speech sound disorders [59]. Regarding inconsistency of speech errors, the participants underwent two tasks, according to which isolated vowel and consonant imitated production skills were evaluated by targeting the sounds /a, i, u, o, m, p, b, t, d^, k, g^/. The PD and TD groups presented the highest scores in both tasks. The administration of S.O.P.S.M task, during which the /a, i, u, o, m/ sounds were targeted, did not portray statistical significance between the aforementioned groups. It was predicted that children with CAS would encounter more difficulty when asked to imitate isolated vowel sounds multiple times and would produce more vowel errors. Data partially supports this hypothesis. Although children with CAS and CAS+PD made more errors on the items during which inconsistent productions of vowels were assessed, compared to the PD and TD children, the error rates were not considered significantly low. Moreover, their performances were close to each other. It was observed that the CAS group obtained a lower score when administered the M.S.O.P.S.M. task, compared to the other participants, during which they were asked to imitate the phonemes /p, b, t, d^, k, g^/ multiple times. In parallel with Aziz et al. [39] it was found that children with CAS produced fewer accurate isolated consonants than children with CAS+PD and PD. In this respect, the study's findings do not attribute a significant differential value to general imitated isolated speech sound production. Therefore, the in-depth evaluation of severity and specific aspects of inconsistency, rather than the determination of its presence, would be considered more useful in discriminating CAS from PD.

CONCLUSIONS

This study sought to provide information concerning the reliability of quantitative measures, whose combined evaluation could be used to distinguish CAS from PD, by non-English clinicians, in order to determine whether CAS speech characteristics are seen universally. Caution is recommended when drawing diagnostic conclusions about the presence or absence of CAS based on one feature alone, due to the fact that the individual detection of the aforementioned measures is considered inadequate to distinguish between CAS and PD.

According to the results of the study, the three features incorporated in ASHA's report [1] provide direction for differential diagnosis in the Greek language. However future investigations in the Greek language can elucidate the extent to which the exploration and interpretation of the above stated features, can guarantee a CAS diagnosis or if the implementation of a broader set of diagnostic criteria, such as the Mayo checklist [26] adapted in Greek, would be significant in supporting a CAS diagnosis.

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