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Childhood apraxia of speech evaluation in autism spectrum disorders: three clinical cases report

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ABSTRACT

Introduction: Among the communication impairments found in subjects with Autism Spectrum Disorders (ASD), recently the literature has suggested a comorbid relationship with childhood apraxia of speech (CAS). The aim of the present study was to report the CAS assessment of 3 children diagnosed with ASD. **Report:** The subjects were three children aged 4 to 6 years, with a medical diagnosis of ASD. The language development assessment (LDA) was performed in the subjects, as well as the ABFW vocabulary evaluation and oral praxis (verbal, orofacial, a sequence of movements, and parallel movements) and evaluation of vocal, prosodic, and speech characteristics. All subjects had moderate language delay with better performance in the receptive area. Difficulties in oral praxis tasks were more evident in one of the subjects. Vocal, prosodic and speech features of all the cases were compatible with CAS. **Conclusion:** In the 3 cases reported, CAS signs were identified with impaired oral motor skills, prosody, and oral praxis, as well as inconsistent speech sound production.

Keywords: speech, language and hearing sciences; autistic disorder; apraxias; speech; diagnosis.

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INTRODUCTION

Autism Spectrum Disorders (ASD) are characterized by being a neurodevelopmental disorder with impairments in social communication, social interaction, presence of repetitive movements, and restricted interests[1]. Among the communication difficulties found in cases of ASD, recently, the literature has pointed out a comorbidity relationship with childhood apraxia of speech (CAS)[2-5], with emphasis on a study[4] that found that 63% of children initially diagnosed with ASD also had CAS.

According to the American Speech Language Hearing Association (ASHA)[6], childhood apraxia of speech (CAS) is a neurological disorder in which the planning and programming of movement sequences of speech sounds are impaired in the absence of neuromuscular deficits[6]. Such difficulties produce symptoms such as inconsistency in speech production with phonemes, syllables, words, and phrases, difficulty in sequencing syllables, as well as inappropriate prosody, especially in performing the lexical or phrasal accent[6].

Despite all the advances in research on CAS, the difficulty in differentiating it from other speech and language disorders still exists[7-10]. In the absence of a standardized

assessment instrument that has reliable psychometric measures that can indicate markers that safely differentiate CAS from other language disorders, the current gold standard is the observation of an experienced clinician[8,11]. Most of the existing instruments lead the clinician to verify the presence or absence of certain linguistic, speech, and oral motor characteristics, but they do not include an explicit definition of the resources or methods to determine how much or how often each characteristic should be observed[7,8]. At the same time, studies have shown a variety of symptoms, with different degrees of severity and comorbidities that bring clinical challenges to the therapeutic planning of these children[8,12,13].

In the case of children with ASD, the difficulties of diagnosing co-occurrence of CAS are even greater[2-5,13]. In a study[4] that investigated the comorbidity of these two clinical conditions, the authors suggest that these diagnoses may be associated, and young children diagnosed with ASD and other speech difficulties should also be assessed for CAS, just as children with CAS should be assessed for ASD[4].

The results of studies on the subject point to the presence of speech errors, mainly distortions, unusual constructions, changes in prosody, alteration in tone, slow speech and voice, especially breathy in cases of ASD with the need for more in-depth studies and with greater control of variables to investigate this co-occurrence[2-5,13].

It can be considered that the literature on the subject has been growing in recent years, but it is still limited[2-5,11]. In addition, it is worth highlighting a recent study that warns of a possible "overdiagnosis" of CAS in complex neurodevelopmental disorders, in which ASD would be included[13]. The authors state that it is necessary to differentiate cases of CAS from other motor speech disorders in these cases[13].

The current scenario regarding studies on CAS in cases of ASD exposed need more in-depth studies on the topic and make it evident that scientific production is essentially international, which motivated the design of this study, case report, even with its limitations it. In other words, in Brazil, we have little information on CAS assessment procedures in subjects with ASD, which places the design of the present study as an important contribution in the development of new research that can support the assessment and consequently the treatment of these cases, as they point out some studies[4,11].

In this context, the objective of the present study is to report the assessment of apraxia of speech in childhood (CAS) in 3 children diagnosed with ASD.

REPORT

The data were collected according to the norms and regulatory guidelines for research involving human beings. The study was approved by the Ethics and Research Committee (process 2,525,338) and the institution involved. The informed consent form was signed by the patients responsible.

Subjects

Research subjects were three children, aged between 4 years and 1 month and 6 years and 6 months, who were diagnosed with Autism Spectrum Disorder by the same neuropediatrician with classification in ICD-10 F.84 and DSM-V 299.0. They were selected, by observing performance throughout the therapies, because they have signs that could be suggestive of praxis difficulties.

All subjects were verbal, attended kindergarten in a regular school in the private network of the municipality in which they lived, were patients of a private treatment institution, and were evaluated in October and November 2017 by the main researcher.

Initially, an interview was conducted with the parents of each child so that the research could be explained, with information about the procedures and objectives, and the consent and signature of the informed consent were requested.

The inclusion criteria were: 1) absence of genetic, metabolic, visual, and/or auditory alterations (information collected in medical reports from the institution's medical records, without mentioning the tests performed); 2) IQ score (intelligence quotient) equal to or greater than 70 in all children, verified by means of an evaluation made by the same neuropsychologist using the WISC instruments (Wechsler intelligence scale for children), SON-R (non-verbal test of intelligence revised), psychometric properties of the revised psychoeducational profile (PEP-R); 3) absence of bilingualism at home and school in all cases.

All subjects were submitted to three procedures. 1) Language assessment performed by the language development assessment (LDA) tests and vocabulary test of the ABFW test for children's language[14]. 2) Evaluation of oral praxis: performed by the researcher in a single and individual session, using the instrument by Bearzotti et al.[15] (in free translation) consisting of isolated, sonorized, sequential, and parallel movement tasks. 3) Evaluation of vocal, prosodic, and speech characteristics: performed in a single and individual session, following the methodology described in the study by Shriberg et al.[2], with two adjustments: in the original study, the evaluation was carried out with parents and with a child in a situation of spontaneous speech, being filmed. A random stretch of approximately 5 minutes was taken from this material to analyze the child's performance, to determine the presence of CAS. In the present study, the procedure was developed in a playful interactive scene between the researcher and each of the children, filmed and later transcribed. A 5-minute random cut of the situation was made, and this material was analyzed according to the categories established by Shriberg et al.[2] with regard to voice, prosody, and speech.

In the original study, software that was not adapted to Brazilian Portuguese was used. Thus, the present study sought

to establish categories of analysis exactly like those used by researchers: a) voice: loudness, resonance, pitch and voice quality; b) prosody: intonation, speed, and vocabulary and phrasal accent; c) speech: substitution, omission, phonetic distortion, and unusual constructions.

Results

The presentation of the cases is described in Table 1. It is possible to verify that the three subjects presented moderate language delay with better performance in the receptive area according to the LDA test. However, in the vocabulary test of the ABFW test, case 1 presented greater lexical difficulty, with a higher rate of non-designations (60%). Cases 2 and 3, on the other hand, presented usual designation rates of 80% and 60%, respectively.

For better visualization of the results, the evaluations of oral praxis (voiced, orofacial, sequence of movements, and parallel movements) and the evaluations of voice, prosody, and speech are presented in Tables 2 and 3, respectively. In the description below, it is possible to monitor the performance of each subject in the referred assessments.

Case 1: Age 4 years and 1 month. Regarding the sound praxis: of the 12 commands, 3 were performed after the request, 6 were performed after imitation, and 3 were not performed. Regarding the 12 movements of orofacial praxis, 1 was performed after the request, 8 were performed after imitation, and 3 were not performed. In the evaluation of the 6 movement sequences, case 1 performed only 2 sequences after imitation. The same difficulty was verified in the performance of parallel movements, in which only 1 of the movements was performed after being requested. Regarding the voice, prosody, and speech evaluations, in case 1, a breathy voice was observed, with a sharp pitch and balanced resonance; prosody with speed and vocabulary tone altered by segmentation operated in verbal production. There was also, in its speech, posteriorization in the production of phonemes, as well as omissions and substitutions of oral phonemes for nasal ones.

Case 2: Age 5 years and 2 months. In the evaluation of the sound praxis, case 2 performed 9 commands after request, 2 after imitation, and only 1 was not performed. The movements of orofacial praxis, 6 were performed after request, 5

Table 1: Presentation of cases

Case	Sex	Age	Age at diagnosis	ABFW test – Vocabulary	LDA test	WISC, SON-R, PEP-R, IQ
1	Female	4 years 1 months	3 years	highest rate of non-designations (60%)	moderate language delay with better performance in receptive area	94
2	Female	5 years 2 months	2 years 6 months	highest index of usual designations (80%)	moderate language delay with better performance in receptive area	136
3	Female	6 years 6 months	2 years	reasonable rate of usual designations (60%), with replacement processes (30%).	moderate language delay with better performance in receptive area	81

ABFW test: children's language test, performed only the vocabulary area; LDA test: language development assessment; WISC, SON-R, PEP-R, IQ: scales for intelligence assessment.

Table 2: Oral praxis of subjects.

Subject	Sound praxis (n=12)			Orofacial praxis (n=12)			Sequence of Movements (n=6)			Parallel Movements (n=5)		
	Solicitation	Imitation	Absent	Solicitation	Imitation	Absent	Solicitation	Imitation	Absent	Solicitation	Imitation	Absent
1	3	6	3	1	8	3	0	2	4	1	0	4
2	9	2	1	6	5	1	3	2	1	5	0	0
3	7	4	1	7	5	0	0	6	0	2	3	0

Table 3: Voice, prosody and speech of subjects.

Subject	Voice			Prosody				Speak		
	Voice quality	Resonance	Pitch	Loudness	Velocity	Segmentation	Tonicity	Replacement	Omission	Distortions
1	Blowing	Equilibrated	Acute	Adequate	Slow	Choppy	Changed	Oral by nasal, posteriorization, deaf by audible, occlusion	Initial consonants, /p/, /m/	Absent
2	Blowing	Equilibrated	Acute	Adequate	Slow	Choppy	Changed	Anteriorization, nasal occlusive, nasalization	Initial consonants, /m/, /l/	Absent
3	Blowing	Equilibrated	Acute	Weak	Slow	Choppy	Changed	Occlusion	Initial consonants and consonant groups	Absent

after imitation, and 1 was not performed. Of the 6 sequences of movements evaluated, 3 were performed after request, 2 after imitation, and 1 was not performed. In the evaluation of parallel movements, case 2 was able to perform all the requested movements. Regarding the voice, prosody, and speech evaluations, there was a breathy voice with a sharp pitch and balanced resonance; prosody with speed and vocabulary tone altered with broken segmentation. Case 2 also presented phonemic substitutions and omissions in its speech.

Case 3: Age 6 years and 6 months. Performed 7 of the commands of sound praxis after request, 4 after imitation, failing to perform only 1 of the commands. Of the 12 orofacial praxis, it performed 7 after the request and 5 after imitation. The sequences of movements were performed after imitation, and parallel movements were performed 2 after request and 3 after imitation. The voice, prosody, and speech evaluations showed a breathy voice with a sharp pitch and balanced resonance. Regarding prosody, there was weak loudness, slowed speed, broken segmentation, and altered tone. In the speech, phonemic substitutions and omissions were observed, with no evidence of distortion processes.

DISCUSSION

The aim of the present study was to report the evaluation of CAS in 3 children diagnosed with ASD based on the combination of 2 methodological procedures that are based on the observation of the patient's performance in oral praxis tests and the analysis of vocal, prosodic vocal characteristics and speech, considering that the current standard for diagnosing CAS is a clinical observation to verify the presence or absence of these characteristics, without any operational definition or standardized instruments[7,8,11].

From the evaluations carried out, it was found that the 3 subjects presented CAS, with impaired oral motor skills, prosody, and oral praxis, as well as inconsistency in the production of speech sounds. Such characteristics are the most frequently reported by clinicians and researchers[6-8,10,11] as being necessary for a differential diagnosis between CAS and other speech sound disorders since the inconsistency in speech is not enough to justify an CAS diagnosis[6].

It was observed that case 1 presented a greater impairment in oral praxis skills, using the imitation feature so that some tasks could be performed with the initial request for commands being insufficient. Although it is possible to assume that the performance in the vocabulary test would have implications for her ability to understand what could justify this result in the initial requests, it was found that the researcher's model for the imitation to be performed served as support for the accomplishment of just some of the sound praxis, as well as just some of the orofacial praxis. Such result indicates unsatisfactory performance in the evaluated oral praxis skills. Itis in agreement with what we find in the literature about the evaluation procedures of the CAS that use not only verbal requests but also are based on imitations as widely exposed in the systematic review on this subject by Gubiani et al.[7].

It was also observed that the subject in question had greater difficulties in tasks that required more complex orofacial praxis skills, such as requests for movement sequences or parallel movements. Here, it is worth noting not only the praxis skills required for these 2 tests but also the issues of memory, social and interaction, difficulties inherent to the frameworks of ASD[1,3]. In this direction, we found a study[9] on cognitive functions in CAS whose results indicate that children with this diagnosis have a poor performance in sensory-motor functions and sequential memory[9].

In cases 2 and 3, better performance was observed in assessments of oral praxis, despite presenting vocal, prosodic changes, and speech inconsistencies in the assessment of spontaneous speech substrate, as well as case 1.

In the vocal assessment, all children had the same performance: breathy voice, heightened with balanced resonance. Only Case 3 presented weak loudness. In the evaluation of prosody, it was observed in all children, speed and vocabulary tonicity altered by the segmentation operated in verbal production, also compromising the rhythm and the elaboration of prosodic curves, which is in accordance with what we find in the literature on CAS[2,4,6,8,10,12,13].

In the assessment of speech, what stands out most in the results are the unusual constructions, as well as the unsystematic and heterogeneity of the substitutions operated by the children, indicating a certain articulatory inaccuracy, findings similar to those described in the literature[2,10,13].

Although the reported cases present difficulties compatible with a diagnosis of CAS with impaired oral motor skills, prosody, voice, and praxis, we cannot fail to highlight the complexity involved in the investigation of CAS in cases of ASD, signaled by several studies[2-5,13] including the one on which our methodological proposal is based by Shriberg et al.[2].

In this direction, it is necessary to point out the limitations of the study design presented here. First, in relation to the sample limitation and the impossibility of generalizations based on the findings exposed here. Second, limitations on the diagnostic determination of ASD. In this sense, it is evident the need for studies that investigate in greater depth and with a larger sample the relevance of using the methodological resources used here.

It is worth emphasizing that studies on CAS in cases of ASD are important and necessary since the idea of comorbidity has been advocated. Still, assessment and therapy, that is, how to detect and treat praxis difficulties in these cases, if confirmed by research, are still little investigated which has important clinical implications.

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