

GLOBAL JOURNAL OF MEDICAL RESEARCH: K INTERDISCIPLINARY Volume 18 Issue 7 Version 1.0 Year 2018 Type: Double Blind Peer Reviewed International Research Journal Publisher: Global Journals Online ISSN: 2249-4618 & Print ISSN: 0975-5888

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By Marlene Escher Boger, Juliana Siqueira, Cleybson Araujo, Alex Maciel & Denise Torquato

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Results: When evaluating the function of outer hair cells in children with ASD, the findings reveal that 55.6% of the children presented alterations in the functioning of the outer hair cells of the cochlea.

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GJMR-K Classification: NLMC Code: WL 705



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Distortion Product Otoacoustic Emissions in Children with Autism Spectrum Disorder

Marlene Escher Boger ^a, Juliana Siqueira ^o, Cleybson Araujo ^e, Alex Maciel ^w & Denise Torquato [¥]

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Conclusion: The DPOAE test proved to be efficient for evaluating children with ASD, but the need to use other tests to determine a safe diagnosis is recognized.

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I. INTRODUCTION

utism Spectrum Disorder (ASD) is a complex behavioral syndrome with multiple causes that combine genetic and environmental factors which cause alterations on the motor, psychological and neurological development, thus, jeopardizing the language, the cognition and social communication of the individual¹⁻². Besides, other characteristics such as stereotypical movements. variable intelligence standards and vulnerability can also be observed³. ASD manifests prematurely before the child is 36 months old⁴⁻⁶. Its etiology still hasn't been found and its diagnosis is defined by the set of displayed symptoms^{7,2}.

In the late years, autism has reached a considerable expression. There's a worldwide estimation of 70 cases for 10,000 inhabitants, and the incidence rate seems to be higher for males⁷. In Brazil, about 27.2

cases of autism were reported for 10,000 inhabitants. However, there doesn't seem to be any known association with racial, social, economic or cultural aspects⁸.

As to the linguistic aspects, children diagnosed with ASD manifest alterations on the socioemotional development, displaying difficulty to establish a normal conversation, reduced share of interests, emotions of affection, beside the difficulty to begin or respond to social interactions. They also display difficulty at nonverbal communication employed in social interaction, alternating, for example, between verbal and nonverbal communication. Other characteristics, such as difficulty to develop, maintain and understand relationships, adjust their behavior to fit diverse social contexts and share imaginative games are also observed in children with ASD⁹. Souza and collaborators (2009) consider that the alterations on language in individuals with ASD usually comprise delay or failure on the development of language, not compensated by gestures or mimics; failure in responding to the communication of others; relative failure in beginning or maintaining communication exchange; usage of stereotypical and repetitive language; idiosyncratic usage of words and abnormalities on speech prosody¹⁰.

As to audition, ASD bearers usually display hypersensitivity of hyposensitivity to sound stimulation. Their visual-spatial processing seems to be whole, though¹¹⁻¹². Lately, some studies have evaluated the audition of children ASD, and some controversies have arisen. In 2014, researchers evaluated children with ASD and concluded that the audio logical findings are compatible with normality both at behavioral evaluation and electrophysiological evaluation of audition¹³. In 2016, other researchers identified an alteration on the internal ear of children with ASD that can impact on their ability to recognize speech. The results concluded that the otoacoustic emission (OAE) test can be used to identify children under risk of autism at premature age. That study investigated the audition of children aged between 6 and 17 years old. About half of them were diagnosed with ASD. It was found that children with ASD face difficulty to hear specific frequencies (1-KHz) that are important for the discourse of speech processing. They also found correlation between the cochlear impairment and the gravity of the symptoms of autism¹⁴.

Evaluating the audition of children with ASD is often a challenge. Such evaluation can be done through

Author $\alpha \sigma p$: Speech Therapist, Audiologist, MA and PhD in Health sciences. Professor at UNIPLAN. e-mails: marlene.escher@gmail.com, ju.domingoss@gmail.com, cbn@hotmail.com.br

Author ⓒ ¥: Undergraduate Student in Speech Therapy at UNIPLAN. e-mails: alex-junio2014@hotmail.com, denise.s.silveira@hotmail.com

objective and subjective exams. Patients with ASD, for having difficulties regarding interaction, attention, perception and memory, make the application of subjective exams difficult and, thus, answers may be mistaken for auditory loss, because they depend on the participation of the individual. Thus, objective exams such as the Brainstem Auditory Evoked Potential (BAEP) and Otoacoustic Emissions (OAE) assure more trustable results¹⁵⁻¹⁶.

Otoacoustic emissions are the sound energy captured by the acoustic meatus, produced by the contraction of the external hair cells of the cochlea. They can be either spontaneous or evoked. Evoked OAE are classified in three types that vary according to the stimulus: Transient, Distortion Product and Stimulus-frequency. In this study, we chose to use the Distortion Product Otoacoustic Emissions (DPOAE). It's a quick, painless, non-invasive procedure that evaluates the answers obtained by cochlear hair cells to the simultaneous acoustic stimulation of two pure tones. It can detect cochlear lesions, from the basal coil to the apical coil¹⁷.

Considering the controversies in the literature on the communication abilities of children with autism spectrum disorder, how much damages to the hearing function are related to those difficulties, and the possibility of an objective evaluation of the cochlear function of such subjects, the goal of this study is to evaluate DPOAE in children with Autism Spectrum Disorder.

II. Methods

It's a transversal descriptive study whose participants were children diagnosed with autism spectrum disorder, aged between 4 and 11 years old. The evaluated population was selected through convenience sample and it was composed only by those subjects that accepted to participate in the research. Those responsible for the participants were informed about the methodological procedures and signed the Consent Form (Attachment 1) before any procedure was conducted.

The data collection was conducted at Clínica Escola de Fonoaudiologia do Centro Universitário Planalto do Distrito Federal (Uniplan), and the sample comprised patients that attend the Supervised Attendance of Child Language of that Institution, diagnose with ASD.

In order to select that sample, the following exclusion criteria were previously defined: presence of other associated impairments, diagnosed by an expert team (psychiatrist and neurologist, speech therapists and psychologists), audio logical risk factors, such as prematureness and usage of ototoxic medicine, and presence of alterations on the middle ear, observed in meatoscopy.

All those responsible for the children answered to the specific audio logical anamnesis (Attachment 2). The audio logical evaluation began with the otoscopic inspection with the purpose of discarding the presence of structural anomalies and/or the obstruction of the external acoustic meatus that might hinder the conduction of the audio logical exam. Then, the analysis of the distortion product otoacoustic emissions (DPOAE) with the purpose of evaluating the cochlear functioning, more specifically the external hair cells. The Oto Read Screener equipment of the INTERACOUSTICS brand was used. DPOAE were evaluated through the simultaneous presentation of two different pure tones (F1 and F2), expressed through the reason of 1.22. The intensity parameter L1=65 and L2=55 dB was used, and the cochlear conditions were measured in the frequency of 2 kHz, 3 kHz, 4 kHz and 5kHz. The analyses of DPOAE were conducted through frequency, following the amplitude criteria (SD) higher than -5 dB and the relation between signal/noise (SN) higher than 6 dB. The occurrence of answers to DPOAE on a frequency was considered when the values established in the two foresaid criteria were observed on that frequency. Those that got answers for at least 3 out of the 4 tested frequencies were considered the normality standard for external hair cells functioning, for both the amplitude criterion and the signal-to-noise ratio criterion.

The collected exams were printed during the test. This enables the visualization of the amplitude and signal-to-noise ratio parameters in DPOAE by separate frequencies.

Those responsible for the children were communicated about the result of the exam immediately after its conduction. In case of alteration of the test, they were guided to the complementary Otorhinolaryngological evaluation and BAEP.

The collected data was transported to electronic spreadsheets, through which they got analytical treatment, both central tendency statistics (mode, mean and median) and variance (standard deviation). They were also presented in diagrams and tables, through the usage of the Excel tool.

This work was submitted and approved by the Ethics in Researches with Human Beings Committee at Faculdade Integrada da União Educacional do Planalto Central - FACIPLAC, with register number CAAE 70136017.0.0000.5058 (Attachment 3).

III. Results

First, 17 otoacoustic emission exams were scheduled preceded by meatoscopy and anamnesis. 5 out of this didn't attend, and 3 didn't collaborate for the test conduction, making the evaluation conclusion impossible. By the end, there were 9 children with ASD evaluated in the DPOAE test. The evaluated subjects had an average age of 6.5 years (SD \pm 2.4). Regarding gender, 100% of the children were male (Table 1).

Table 1: Characterization of the Sample according to					
Age and Gender					

Characteristics		Descriptive Statistics		
	Mean ± SD	$6,5 \pm 2,4$		
400	Median	6		
Age	Minimum	4		
	Maximum	11		
Gender		Ν	%	
Male		9	100	
Female		0	0	

 $\pm SD$ - standard deviation

According to the results obtained in PDOAE, it was observed that 5 out of the 9 evaluated individuals (55.6%) obtained altered results and 4 are within the normality standards. Out of those altered results, only 1 (11.1%) presented alterations in both ears and in all tested frequencies. When each ear was evaluated separately, that variable displayed unilateral alterations of 33.3% on the left ear and 11.1% on the right ear, characterizing failure on the functioning of the external hair cells of the cochlea (Table 2).

Table 2: Prevalence of PDOAE according to Laterality.

PDOAE Prevalence				
	Normal		Alter	ed
Laterality	Ν	%	Ν	%
BE	4	44,4	1	11,1
LE	5	55,5	3	33.3
RE	7	77,7	1	11,1

BE - both ears; LE - left ear; RE - right ear

As to the PDOAE analysis criterion, only 1 individual presented alterations in more than one of the frequencies tested in the signal-to-noise ratio criterion. In the amplitude criterion, 5 individuals displayed alterations on the functioning of the external hair cells, and 4 obtained answers within the normality in both criteria (Table 3).

Table 3: Alterations in PDOAE according to the Criterion Employed

PDOAE Prevalence						
	Nor	mal	Altered		Total	
Criterion	Ν	%	Ν	%	Ν	%
Amplitude	4	44,4	5	55,6	9	100
Signal / Noise	8	88,9	1	11,1	9	100

When PDOAE were analyzed regarding the average amplitudes, the laterality of the ears and the evaluated frequencies, it was observed that, with the increase of frequencies, there is a decrease of amplitudes in the right ear. As to the left ear, the frequency with the highest amplitude is 3kHz (3.3dB)

and the lowest amplitude is on the frequency of 5KHz, which result is lower than -5dB (-6.2dB), showing that the alteration of external hair cells in children with ASD is mainly on that frequency (Table 4).

Table 4: Overall Mean of the DPOAE Amplitudes according to the Laterality (Left and Right Ears) and the Evaluated Frequencies

PDOAE Amplitude				
	R	E	LE	
Freq. (kHz)	Mean	±SD	Mean	±SD
2	3,7	8,1	1,9	9,4
3	0,0	10,2	3,3	5,2
4	-1,4	9,3	-2,8	9,4
5	-2,7	7,0	-6,2	10,5

 \pm SD - standard deviation

Regarding the DPOAE means in the signal-tonoise ratio, the laterality and evaluated frequencies, it was verified that, in both ears and in all the frequencies, the means area higher than 6 dB, a normality criterion (Table 5).

Table 5: Overall Mean of the Signal-To-Noise Ratio of the PDOAE according to the Laterality (Right Ear) and Evaluated Frequencies.

PDOAE Signal-To-Noise Ratio				
	RE	Ξ	L	E
Freq.(kHz)	Mean	±SD	Mean	±SD
2	13,0	6,0	8,2	10,0
3	17,3	8,9	13,7	6,6
4	16,6	9,2	12,9	11,5
5	15,2	8,1	11,2	10,1

 \pm SD - standard deviation

IV. DISCUSSION

In this chapter, the results found in the study will be interpreted, and compared to the data of the authors mentioned in the literature revision, whenever it's possible.

The possibility of identifying a cochlear alteration in children with autism spectrum disorder motivated this study, and it motivated several scientists to research through the OAE test. The base of the selection of the distortion products for this research was the possibility of evaluating the cochlear activity in specific frequencies, providing a wide analysis compared to the transient emissions that evaluate the cochlea globally. For the conduction of this test, it's essential that the middle ear is in proper physiological conditions. It is an efficient, quick and objective exam for the differential diagnosis and the monitoring of the audition. However, the Otoacoustic Emissions test is considered a complementary evaluation, and it alone can't diagnose auditory losses. So, complementary behavioral, electroacoustic and electrophysiological audition exams must be requested.

As to the characterization of the sample, all the evaluated children were male, supporting Volkmar and McPartland (2014), who assert that the incidence rate of OAE seems to be higher for the male gender⁷.

Regarding the auditory characteristics, it was verified that almost half (55.6%) the children displayed altered DPOAE according to the established evaluation criteria, which differs from the study conducted by Romero and collaborators (2014), in which the findings displayed auditory normality in every conducted test, including the otoacoustic e missions¹³.

Besides the prevalence registers, the occurrence of OAE is usually analyzed by a set of criteria. In this research, the amplitude and signal-tonoise ratio criteria, with the purpose of evaluating the occurrence of DPOAE in children with ASD. In this study, the selection of amplitude criterion higher than -5 dB and the signal-to-noise ratio criterion higher than 6 dB proved to be efficient in the detection of alterations of external hair cells in children with ASD. We know that in the clinical practice, some researchers employ more rigid criteria, such as signal-to-noise ratio higher than 8 dB or the absence of answers on a single frequency would be considered an alteration¹⁸. The adoption of these criteria in this study would raise the possibility of even worse results.

When you compare the amplitudes and signalto-noise ratio amplitudes, you verify that the worst results refer to the amplitude criterion. This data shows that the selection of analysis criteria area extremely important to determine the normality or the alteration of the external hair cells. Gorga and collaborators (1997) assert that the DPOAE amplitude shows a decrease with aggravation of the auditory threshold in individuals with cochlear auditory loss. Thus, the amplitude evinces the real functioning of the cochlea. Therefore, it is an indispensable criterion. Some OAE equipment are already factory-set to consider normality, based only on the signal-to-noise ratio criterion. In this study, we remark that the usage of only one criterion is not enough to establish normality or alteration of the cochlear activity.

We must remark that only the observation of the signal-to-noise ratio of the data presented in this work would raise the percentage of normality of the evaluated children to 88.9%, while with the combination of criteria, signal-to-noise ratio and amplitude, such percentage of normality usually decreases to 44.4%.

When the amplitudes means were analyzed separately, it was observed that the frequency of 5KHz showed the worst means, thus differing with the study of Bennetto and collaborators (2016), which showed that children with ASD have difficulty to hear specific frequencies in the range of 1-2KHz14. On the other

hand, it supports such research, when it asserts that the OAE test can be used to identify alterations in

One of the aspects that stood out the most in this study was the occurrence of the only individual that presented alterations in both ears, in both criteria, and in every frequency. At the age of two, the child had been evaluated with BAEP and OAE during the phase of diagnostic investigation of ASD, which result, then, was within the normality standards. Currently, at the age of 5. when he was invited to participate in this study, he was evaluated with DPOAE and didn't show cochlear answers. So, he was guided to the conduction of a new BAEP on which severe bilateral auditory loss was confirmed. Such fact evinces the possibility of late auditory loss. Thus, it reinforces the importance of the annual auditory monitoring in children with ASD with the purpose of preventing that late auditory alterations appear and don't be detected, contributing even more for all the difficulties already found in language for that population.

Considering these observations, and admitting the need to advance the studies and researches in this field, we suggest the development of new studies, with the purpose of establish the audiological profile of individuals with ASD, as the suggestion to conduct a case-control study, comparing the audiological profile of children with ASD with children with no complaints, as well as the conduction of complementary exams, such as transient evoked otoacoustic emissions, BAEP and tympanometry, the conduction of longitudinal studies and the study with the highest number of tested frequencies.

V. CONCLUSION

The DPOAE test proved to be efficient to evaluate children with ASD, but we recognize the need to conduct other tests to determine a safe diagnosis.

Conflict of Interests

There's no conflict of interests.

Finance

There was no financing.

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