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Role of Otoacoustic Emissions in Hearing Assessment of Children with Autism

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Abstract

Evaluation of hearing is essential in the diagnostic process of children suspected of having autism spectrum disorder (ASD). The study aimed to examine the efficiency and feasibility of utilizing otoacoustic emissions as an objective test to assess hearing in children diagnosed with ASD. A pilot study was conducted over a 3-year period (from September 2019 to November 2022) in the Audiology Department at the Center for Specialty Surgeries, Benghazi, Libya. A total of 61 children with autism aged between 2 and 15 years, referred for hearing evaluation, were enrolled in the study. The clinical procedure consisted of an otoscopic ear examination and Otoacoustic Emission (OAE) recording. OAE testing was attempted on the study sample (n 51) who met the inclusion criteria and completed successfully for 44 participants (86%): 40 cases (91%) from the first attempt, while 4 cases (9%) needed a second or more visit. Four autistic children (9%) had absent otoacoustic emissions in one ear. Testing could not be completed for 7 children (14%) due to behavioral difficulties. The results of this study showed the feasibility of using Distortion Product Otoacoustic Emissions (DPOAEs) as an objective tool in the initial assessment of hearing for children with autism, for whom obtaining reliable behavioral responses can be challenging.

Keywords: Autism, Autism Spectrum Disorder, Children, Hearing, Otoacoustic Emissions.

INTRODUCTION

Autism spectrum disorder (ASD) is defined by the American Psychiatric Association's Diagnostic Statistical Manual of Mental Disorders, Fifth Edition, Text Revision (DSM-5-TR) as a neurodevelopmental disorder characterized by lifelong challenges in social communication and interaction, in addition to the presence of restricted interests and repetitive behaviors (American Psychiatric Association, 2022).

Globally, the prevalence of autism has noticeably increased over time. It is estimated that about one in 100 children has autism spectrum disorder, with a significant male predominance (Zeidan et al., 2022; WHO, 2022; Maenner et al., 2023). In the United States, according to the 2020 Centers for

Disease Control and Prevention (CDC) Surveillance Report, ASD prevalence was one in 36 per 1000 children aged 8-years (CDC, 2023; Maenner et al., 2023). Based on a systemic review conducted in the Arab Gulf region, the prevalence of autism in the six Gulf Cooperation Council countries ranged from 1.4 to 29 per 10,000 persons (Salhia et al., 2014). In Libya, the prevalence varied between one in 190 and one in 178 per 1000 children in 2011 and 2015, respectively (Zeglam et al., 2012a; Zeglam et al., 2012b; Zeglam et al., 2016).

Hearing conditions in children with autism spectrum disorder are variable. Some children with ASD don't exhibit any hearing difficulties. While other autistic children have some degree of auditory abnormalities, such as hypersensitivity to sounds, difficulty hearing in background noise, unresponsiveness to certain verbal stimuli, hearing loss, and tinnitus (Danesh et al., 2015; Dayem et al., 2018). The co-existence of hearing loss and ASD was reported in the literature (Rosenhall et al., 1999; Roper et al., 2003). Gallaudet Research Institute survey (2009-2010) revealed that up to 40% of hearing-impaired additional have children comorbidity, including visual impairment, learning disabilities, cerebral palsy, and autism (Szymanski et al., 2012). Some research claimed a higher prevalence of ASD among hearing-impaired children than in normal-hearing children, while other studies indicated no difference between the ASD population and the general pediatric population (Beers et al., 2014). It is estimated that one in 59 children with hearing loss is also diagnosed with ASD (Szymanski et al., 2012). Results of the Zeglam et al. (2016) study conducted at Al-Khadra Teaching Hospital in Tripoli revealed a prevalence of 11 per 1000 (1.1%) of sensorineural hearing loss among Libyan children with ASD.

Evaluation of hearing is essential in the diagnostic process of children suspected of having ASD who presented with early communication difficulties and or delayed language development (Tas et al., 2007; Li et al., 2021). Co-occurrence of hearing loss and ASD can significantly delay the diagnosis of either condition (Mandell et al., 2005; Fitzpatrick et al., 2014; Danesh et al., 2015). Moreover, it is probable that children with hearing loss are being misdiagnosed with ASD (Fitzpatrick et al., 2014). Furthermore, previous research identified several common risk factors between hearing loss and ASD, for example, prematurity, low birth weight, hypoxia, and viral infections (Limperopoulos et al., 2008; Danesh et al., 2015; Allen et al., 2020).

Reliability of hearing thresholds obtained by behavioral audiometry for children with ASD can be poor due to the atypical behavior some ASD children may display during conventional audiological testing, such as limited ability to communicate, uncooperativeness, inability to follow test instructions, dislike wearing headphones, or anxiety to test booth or examiner (Schafer, 2021). Therefore, objective hearing measurement tools, such as otoacoustic emissions (OAEs) and auditory brainstem response (ABR), may be considered valuable in the assessment of the auditory system function of children with autism (Tas et al., 2007; Beers et al., 2014; Bennetto et al., 2017).

Otoacoustic emissions are commonly used in the hearing assessment of difficult-to-test patients who cannot provide reliable behavioral responses (Demopoulos 2016). OAEs are also used to screen hearing in neonates and test for functional hearing loss. Measurement of OAEs in children with developmental disabilities has numerous advantages, namely easy to record, non-invasive, doesn't require behavioral cooperation or response, and provides information about cochlear status in a short time.

The presence of OAEs indicates normally functioning outer hair cells which correspond with essentially normal hearing sensitivity. It also denotes the proper function of the external and middle ear conductive mechanism of hearing. However, OAE testing has some disadvantages; it is very sensi-

tive to noise, greatly affects the middle ear status, can't be used to estimate the degree of hearing loss, and doesn't evaluate neural auditory pathways (Tas et al., 2007; Cunningham, 2011; Kaf et al., 2013; Al-Meqbel, 2016).

Unlike OAEs, estimating hearing thresholds by Auditory Brainstem Response (ABR) may not be possible without sedation for autistic children at an average age of diagnosis, over 3 years old (CDC, 2019; van't Hof et al., 2021). Additionally, running ABR is time-consuming in comparison to recording OAEs. Although ABR is considered the accepted method for predicting hearing thresholds in suspected ASD children (Elmawgoud et al., 2017; Kamita et al., 2020), the previously mentioned drawbacks render ABR unnecessary for assessing the hearing of ASD children without any parental concern of hearing problems especially in developing countries like Libya where shortage of hearing healthcare professionals and cost and availability of medical equipments and supplies are major barriers and constraints (Zeglam et al., 2016).

The present study had two main aims. First, to examine the efficiency and feasibility of utilizing Distortion Product Otoacoustic Emissions (DPOAEs) as an objective initial test to evaluate hearing in children diagnosed with ASD and to help identify those who need comprehensive audiological assessment. It was expected that unsedated OAE tests couldn't be successfully completed in the autism pediatric population. Second, to compare the emission responses of children with autism spectrum disorder and normally developing peers. It was hypothesized that levels of OAEs of children with ASD would be significantly higher than of normally developing children.

MATERIALS AND METHODS

This study was conducted at the Audiology Department at The Center for Specialty Surgeries, Benghazi, Libya, which is one of the two main hearing referral centers in eastern and southern Libya. Verbal informed consent was taken from the parents of all participants.

PARTICIPANTS

Autism Group: Initially, a total of 61 children diagnosed with autism aged between 2 and 15 years enrolled in this study, 46 boys and 15 girls (Ratio 3:1). Among them, 10 children with autism were excluded.

Control Group: 23 age-matched normally developing children, 8 males and 15 females (Ratio 8:15) randomly recruited from patient companions/hospital visitors with non-ear, nose, and throat-related problems.

SAMPLING PROCEDURES

Audiological evaluations consisted of asking parents about any concerns about hearing problems, doing otoscopic examinations, and measuring DPOAEs. Clinical data for all participants who met the study inclusion criteria was examined retrospectively:

Inclusion Criteria

- Referral by a psychologist or a special needs and autism center.
- No parental concern of hearing impairment.
- Normal otoscopic examination (no external auditory canal or middle ear pathology)
- DPOAE test results recorded for frequencies: 1500, 2000, 3000, 4000, & 6000 Hz for both ears

Exclusion Criteria

- Uncertain diagnosis of autism or not documented by a specialist.
- Abnormal otoscopic examination (occluded external auditory canal, congested tympanic membrane, or presence of pressure equalizing tubes).
- DPOAE test results were recorded only for one ear.

Diagnostic DPOAE measurements were performed in a quiet room using Interacoustics Eclipse (A/S DK-5610 Assens, Denmark) DPOAE probe for each ear while participants were awake and still. The DPOAEs are elicited by two pure tone stimuli [L1=65 and L2=55 dB sound pressure level (SPL)] at primary frequencies of f1 & f2 at a fixed ratio of f2/f1 = 1.2. The DPOAE response was measured at the distortion product generated at (2f1-f2) for targeted five frequency regions: 1500, 2000, 3000, 4000, & 6000 Hz. The testing time was 60 seconds. The detection of the DPOAEs was based on the amplitude of -5.0 dB or more and being at least 6 dB above the average level of the noise floor (Signal-to-Noise Ratio (SNR) = ≥6 dB).

Statistical Analysis

The recorded distortion product otoacoustic emission responses (Distortion Product (DP) level and SNR) of children with autism were investigated and analyzed using Apple Mac Numbers® basic descriptive statistics and compared with those of non-autistic control subjects. The variables from the two groups were compared for the right and left ears separately and both ears together using a two-sample t-test. The level of significance was set at $p<.05$.

Table: (1). Demographic Characteristics of Study Subjects

Variable	Autism Group (n=51)	Control Group (n=23)
Age range, years	2 - 15	2 -12
Age mean, years±SD	6.3±3.39	6.5±2.52
Age median, years	6	6
Gender, n (%)		
Male	40(78%)	8(35%)
Female	11(22%)	15(65%)
Address, n (%)		
Benghazi	44(86%)	18(78%)
Outside Benghazi	7(14%)	5(22%)

SD: Standard Deviation

RESULTS

Table 1 summarizes the participants' demographic characteristics. DPOAE testing was successfully completed for 44 ASD participants (86%), 40 cases (91%) from the first attempt, while the other 4 cases (9%) had to be retested. DPOAE testing couldn't be completed for 7 (14%) ASD children due to uncooperativeness and high noise levels. DPOAEs were present in both ears in 40 (91%) ASD participants, whereas it was present in just one ear in the remaining 4 (9%). Since all children in the control group cooperated, DPOAEs were measured from the first trial and were absent in only one out of 46 ears. Figure 1 displays the average time in seconds taken for DPOAE measurements (probe insertion not included) in each ear for both the autism and the control group. Running the -test revealed no significant difference in time spent to complete the DPOAE test between the two

groups (p= >.05).

There was no statistically significant difference in the DP levels in the right ear and left ear between the autism and control groups (Table 2). For both ears together, the highest DPOAE amplitude was recorded at 1500 Hz, with similar results in both groups.

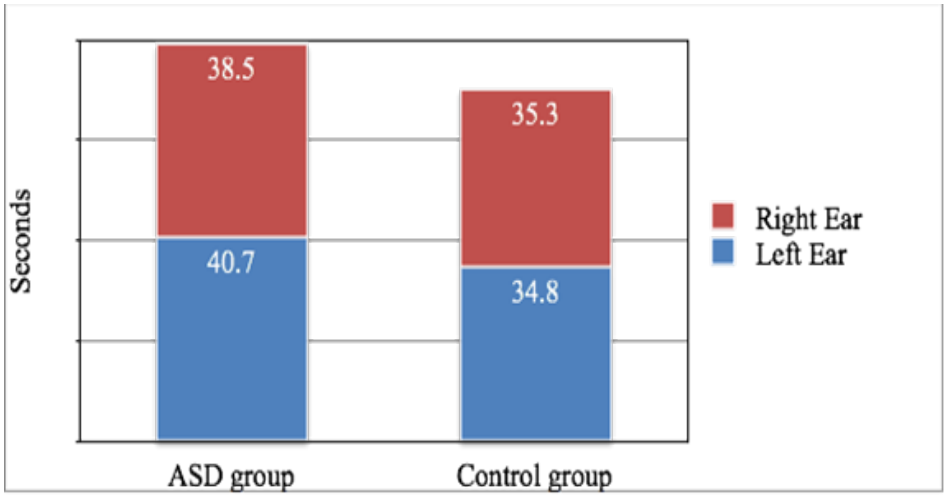

Figure: (1). Average DPOAE test duration in seconds for each ear for both groups

Table: (2). Mean values and standard deviations of DPOAE levels in dB SPL in each ear for both study groups

DPOAE Level (dB SPL)	Right Ear					Left Ear				
Frequency (Hz)	Autism Group		Control Group			Autism Group		Control Group		
	M	SD	M	SD	p-value	M	SD	M	SD	p-value
1500	15.1	7.91	13.64	7.28	.453	13.58	8.16	14.90	5.49	.436
2000	11.42	7.01	12.36	7.34	.613	9.92	9.43	12.15	6.53	.262
3000	7.79	7.05	8.25	6.28	.787	6.38	6.81	8.32	6.69	.269
4000	9.77	6.48	10.53	5.98	.636	8.38	7.82	10.77	5.50	.151
6000	4.19	8.74	4.28	10.60	.974	4.17	8.55	1.65	10.11	.313

DPOAE: Distortion Product Otoacoustic Emission; dB SPL: decibel sound pressure level; Hz: Hertz; M: Mean; SD: Standard Deviation

In regard to signal-to-noise ratio (SNR), the analysis showed significantly higher SNR at 1.5 kHz, 2 kHz, 3 kHz & 4 kHz of the left ears (Figure 2) in the control group than that of the autism group, with the greatest difference observed at 3 kHz (p=<.001). Although there was no significant difference in the right ear SNRs between the two groups, as illustrated in Figure 3, the biggest between-group SNR difference for the right ear was also recorded at 3 kHz. When data from both ears were analyzed together, both groups had the largest combined SNR average at 4 kHz, 19.3 dB in the autism group and 22.3 dB in the control group (table 3). Studying the sex effect on DPOAE responses between 37 males and 7 females in the autism group revealed no significant difference p=>.05.

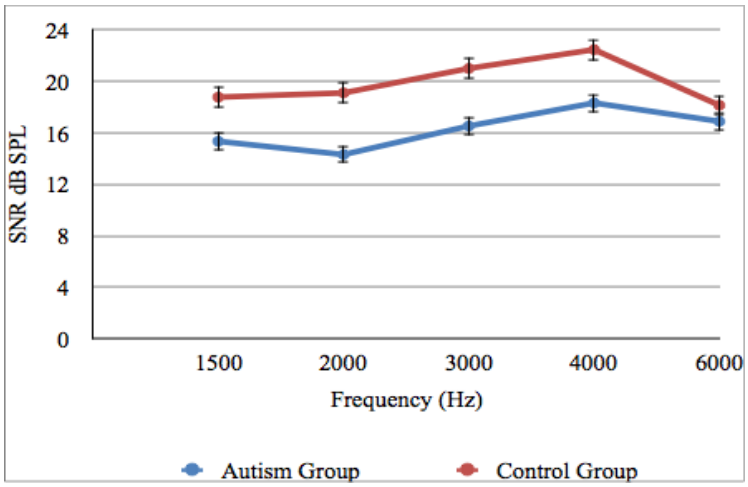

Figure: (2). Comparison of DPOAE SNR at each frequency in left ear between the two groups

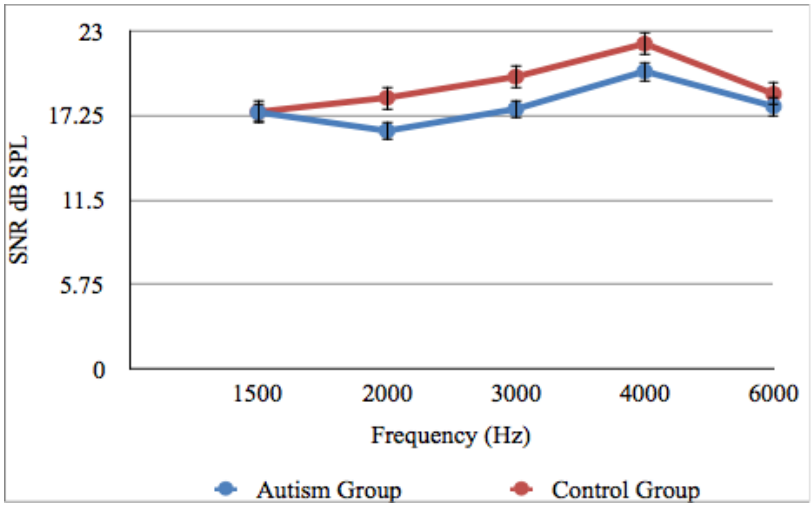

Figure: (3). Comparison of DPOAE SNR at each frequency in right ear between the two group

Table: (3). Mean values of both ears DPOAE SNR at each tested frequency in children with and without autism

Frequency (Hz)	Both ears DPOAE SNR (dB SPL)				p-value
	Autism Group		Control Group		
	M	SD	M	SD	
1500	16.43	5.60	18.17	4.57	.177
2000	15.30	4.88	18.76	4.35	.004**
3000	17.15	4.07	20.48	4.19	.003**
4000	19.33	4.86	22.33	4.23	.011*
6000	17.41	4.92	4.92	5.62	.430

DPOAE: Distortion Product Otoacoustic Emission; SNR: Signal-to-Noise Ratio; dB SPL: decibel sound pressure level; Hz: Hertz; M: Mean; SD: Standard Deviation; *P<.05; **P<.01

DISCUSSION

The lack of cooperation of ASD children reported during audiological testing could be attributed to the sensory sensitivity issues that children with autism are characterized by (Mansour et al., 2021). The abnormal tactile hypersensitivity response to the otoscopic ear examination or the insertion of ear tips of tympanometry or OAE probes into the external ear canal could be the reason behind the high rate of incomplete testing (Andersson et al., 2013; Danesh et al., 2015). In the present study, some strategies for testing hearing in children with autism, as suggested by Brueggeman (2012), were followed, letting the child touch the ear speculum and ear tip, pretending to test a sibling or parent by demonstrating insertion of the ear tips on, or asking parents to practice wearing earphones at home for the few participants who didn't cooperate at the initial appointment. Interestingly, otoscopy was carried out in all autism cases enrolled in the present study. Unlike Kaf, a 2012 study reported a higher percentage of uncompleted otoscopies, whereas DPOAE measurements were obtained without sedation in the majority (86%) of the included autism study sample. Obtaining OAEs under sedation was recommended, especially in severe ASD children, to control motor issues that may interfere with OAE testing (Tas et al., 2017). However, Gungor et al., 2016 observed a significant reduction in DPOAE levels with sedative agents.

As seen in the result section, almost all (92%) study participants had present otoacoustic emissions in both ears, which is suggestive of normal outer hair cell function. A few children (<10% of the autism group and <5% of the non-autism group) had absent emissions in one ear, which is indicative that further evaluation is required to establish diagnosis. Conductive hearing loss due to middle ear disorders is a frequent problem in children with autism, as concluded by Rafal (2016) and Al-Meqbel (2013). Middle ear diseases can impair the transmission of otoacoustic emissions and cause reduced emissions amplitude or absent response (Balatsouras et al., 2012). Tympanometry should be included as an objective tool in the screening of hearing in uncooperative children, particularly autistic children (Rafal et al., 2013; Al-Meqbel, 2016).

Another finding is the higher overall DPOAE SNRs in the control group compared with those in the autism group. A similar finding was confirmed by Danesh & Kaf (2013). While other studies found no difference between the DPOAE SNRs of children with autism and typically developing children (Gravel et al., 2006; Tas et al., 2017). As discussed in the Bennetto et al. (2017) study, the significantly reduced combined DPOAE SNR levels at mid-frequencies without any differences in noise floor may correlate with the speech perception and discrimination challenges persons with autism suffer from. Studying the ear effect on DPOAE SNR responses revealed left ear advantage in the control group. But no significant auditory asymmetry was noted in the autism group, which doesn't agree with what has been mentioned in the literature (Khalfa et al., 2001; Kaf et al., 2013). The inconsistent results could be due to the lack of contralateral suppression of OAEs, as argued in Danesh et al. (2012) study.

This study has several limitations. First, no detailed medical history in the participants' records. Like identifying risk factors for hearing loss, handedness, and severity of autism to compare results accordingly. Second, the inability to get tympanograms for all participants was a restriction, as middle ear status can influence otoacoustic emission response. Third, not measuring DPOAEs below 1500 Hz and above 6000 Hz. Finally, the relatively small sample size for the control group may make it difficult to generalize the outcomes and make them inconclusive.

CONCLUSION

Although no clear evidence exists of a higher risk of hearing loss in children with autism spectrum

disorder, assessment of hearing using objective measurements such as OAE is recommended in children with confirmed and suspected autism. Because the national hearing screening program is not yet routinely implemented on all children in Libya, hence autism spectrum disorder may be identified before hearing loss in those affected by both conditions. Therefore, early identification and timely intervention of hearing impairment in ASD is very important and can reduce the burden of dual disability and improve communication and overall quality of life.

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