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Clinical utility of ABR testing under general anaesthesia in Autistic children

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Abstract

Aim

Hearing assessment in the paediatric population with Autistic Spectrum Disorder (ASD) poses many challenges. A sizeable proportion of these children need general anaesthesia (GA) for an auditory brainstem response (ABR) to confirm hearing or obtain an accurate hearing level. We aimed to investigate the severe hearing loss pick-up rate in our ASD paediatric population who passed the Universal New-born Hearing Screen and evaluate if a general anaesthetic is justified in this population group.

Methods

The medical charts of ASD children with hearing concerns undergoing ABR under GA in our institution were retrospectively investigated for the past 10 years.

Conclusions

Out of the fifty-nine patients, two (3.5%) failed the ABR. One patient was diagnosed with conductive hearing loss and one with sensorineural hearing loss.

Discussion

In our opinion, the pickup rate of severe hearing loss (3.5%) in ASD children is very low. Given the resource-intensive and traumatizing nature of ABR under GA and its potentially damaging effect on these children's cognition, we suggest that there is a great need for the development of an in-clinic hearing assessment modality in this patient group.

Introduction

The prevalence of autistic spectrum disorder (ASD) diagnoses among the paediatric population have increased by 189% between 1996 and 2010.[1,2,3] The same trend was recorded in the Republic of Ireland, with a prevalence of 15 in 1,000 (1.5%).[4,5] This is most likely due to an increase in awareness among medical professionals and community providers involved in caring for children.[6]

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There is debate in the literature regarding the prevalence of hearing loss among the ASD population. It was estimated to be between 1% and 3.5% by previous studies.[2,8] Only 1 in 1,000 ASD children have severe to profound sensorineural hearing loss (SNHL).[7,8,9] Usually, once either ASD or hearing loss is diagnosed, this leads to a delay in the diagnosis of the other condition.[8,10] The severity of autism can also contribute to speech and language development. It is estimated that 25% of ASD patients are nonverbal.[11] ASD patients were also found to have central manifestations, in the form of central auditory processing disorder, although results are still controversial.[3,12,13,14,15] There is no conclusive evidence that children with ASD are at a higher risk of hearing impairment than the general population.[7]

ASD patients often need an auditory brainstem response (ABR) test to evaluate their hearing level as per the American paediatric society guidelines.[8,11] In-office audiological testing in these patients is challenging and behavioural test-retest reliability is poor. Whilst a minority of patients with mild ASD would be suitable for ABR under sedation, a majority of them require testing under general anaesthesia (GA).[16] Visiting the hospital and operating theatre is a stressful and challenging situation for any patient, especially for this group and their families. Unfamiliar places and faces can disrupt their routine which is of great importance to them. Increased use of sedation is reported when dealing with these patients.[17]

It is well demonstrated in the literature the negative long-term effects of GA medication on the brain. Developmental and behavioural delay, processing speed, fine skills, reading difficulties, emotional behaviour, learning, and memory have all been associated, to some degree, with the neurotoxic effect of general anaesthesia agents, especially volatile ones and propofol (a GABAergic agent). These effects are secondary to widespread neuroapoptosis, whereby early exposure to anaesthesia causes long-lasting impairments in neuronal communication and faulty formation of neuronal circuitry.[12,16,17,18] The longer the GA, the bigger the impact on brain development, with researchers finding differences even for procedures over 35 minutes.[17] The Mayo Anaesthesia Safety in Kids study found multiple exposures to GA under age 3 to adversely affect behaviour and learning.[19] The same study showed that the most common procedures performed under this age were otorhinolaryngology (ORL).

Our objective was to investigate the prevalence of severe hearing loss in our ASD paediatric population who passed the Universal Newborn Hearing Screen (UNHS) and evaluates if a general anaesthetic is justified in this population group.

Methods

IMJ
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The ABRs performed between January 2012 and January 2022 in our Audiology department were reviewed. This is a tertiary referral center in paediatric ORL. Since its opening in 1872, the acute national pediatric hospital has cared for 150,000 children per year, including over 45,000 who have attended the emergency department annually.

Inclusion criteria were patients with a diagnosis of ASD who had ABR performed under GA and available data. Their medical and audiological charts were retrospectively reviewed.

ASD was diagnosed by the Early Intervention Group using the Diagnostic and Statistical Manual of Mental Disorder 4th Edition (DSM-4) and DSM-5 criteria.[20] The diagnosis was made pre or post-ABR testing.

Written informed consent was obtained from the parents before the procedure. Premedication was mainly indicated by the parents or team caring for these children. Midazolam, one of the most commonly used benzodiazepines for this purpose was also the most commonly prescribed agent for our patients. It was administered orally, in liquid form by the parent under nurse supervision. General anaesthesia was induced via a mask inhalational agent (sevoflurane) and maintained on an individual basis on volatile, intravenous agents (propofol) or a combination of both. ABR was performed in the operating theatre under GA after examination of the ears, microsuction, and grommet insertion if middle ear fluid was present.

ABR is actually registering (through electrodes placed on the scalp) the neurological activity at the brainstem level after auditory stimulation. This evoked potential is recorded as a waveform, formed by five waves. The ABR was performed using Bio-logic® Navigator PRO Equipment. Electrodes were placed on both mastoid tips and the forehead with bilateral ear inserts. Our institutional ABR protocol is to use a 4KHz tone pip only. If the response was abnormal, the examiner would test 1KHz and if this was abnormal too, bone conduction was performed to confirm SNHL. The testing was performed for one ear followed by testing the contralateral ear in all cases. The pass level was represented by wave V at 40dBnHL corresponding to 30dBeHL (estimated dB HL).[21] All tests were performed by a single senior audiologist.

All the cases were performed as a day procedure. The parents were given the verbal report and a departmental standardized sheet of advice depending on the result, pass or fail.

Approval for our research was obtained from the hospital's Research Committee Group.

Data were collected, processed, and analysed using the Statistical Package for the Social Sciences (SPSS).

Results

IMJ Official Journal of the Irish Medical Organisation

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Out of the one hundred and forty-three ABRs performed under GA, fifty-nine cases (41%) were on ASD patients. Two ASD patients were excluded as they were known to have failed the UNHS (1 CHL, 1 SNHL).

Fifty-seven patients were included in our study. Forty-six were males and eleven were females.

The age at testing ranged from 1 to 8 years, with an average of 3.38 years of age.

The vast majority of our patients were referred by the community paediatricians and general practitioners as outlined in Table 1.

Referral source	**Patients (Nr)**	**Patients (%)**
Paediatrician	18	31.5
General practitioner	14	24.5
Already in our system	13	23
Community audiology	4	7
Not mentioned	3	5
Other ORL services	3	5
Speech and language therapist	2	3.5

Table 1: Referral sources in numbers of cases.

Speech and language delay was the main concern of their parents, with hearing loss being less of a concern, as outlined in Table 2:

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Presenting complain	Patients (Nr)	Patients (%)
Speech delay	49	86
Hearing concerns	8	14
Recurrent acute otitis media	2	3.5

Table 2: Presenting complaints in numbers and percentages.

Seven patients (12%) presented also recurrent episodes of acute otitis media. All these patients were very young (2-3 years old). Only three of these patients had grommets inserted at the time of the ABR testing.

Out of the fifty-seven patients, two (3.5%) failed the ABR testing. One patient had conductive hearing loss (CHL) and one SNHL. Both patients were referred with speech delay and had intraoperative evidence of middle ear effusion.

At the time of ABR, twenty-eight patients (49%) had bilateral dry middle ears and seventeen (30%) bilateral otitis media with effusion (OME). Four cases (7%) had one dry ear and one ear with OME. Nineteen patients (16%) had previous grommets inserted.

Our pickup rate of SNHL in ASD children who passed their UNHS testing is very low (3.5%). None of the patients in our study group was diagnosed with auditory neuropathy disorder.

Discussion

To our knowledge, this is the first published study in Ireland evaluating the clinical utility of ABR testing in the paediatric ASD population. We identified 59 patients who underwent ABR testing under general anaesthetic representing 38% of patients undergoing this procedure at our institution from January 2012 to January 2022. Thus, we were able to assess the diagnostic yield of ABR under general anaesthetic in this population. The male-to-female ratio of four to one in our ASD population is similar to previous studies. [2,3,4]

Behavioural hearing assessments used in typically developing children can be of questionable reliability when used in the paediatric ASD population.[7] The need for communication, interaction, and active involvement in the testing process is impaired by their sensitivity issues.[22] Most of these children have no interest in engaging neither in games or activities nor in responding to sound stimuli. As per the American Academy of Paediatric Position Statement, tone-burst ABR is the best tool available to approximate the

behavioural audiogram in cases where it is not possible to obtain reliable behavioural assessment data.[23]

Indications for ABR were failed in-office behavioural hearing assessments in the setting of speech delay (51% N=29) and parental concerns re hearing loss (72% N=41). This represents the natural disease pattern of ASD with hyporeactivity and decrease interest in sounds but sufficient hearing level. Nineteen percent of our patients are nonverbal, similar to the international published literature.[11] The diagnostic yield of sensorineural hearing loss within our study population was very low at 3.5%.

A third of these children (34%) had evidence of a CHL element (OME) at the time of ABR testing, which is similar to the general population (15-40%).[24] A preoperative in-office tympanometry was not possible in most of these patients due to their sensitivity and behaviour. The median age of 3.37 years at testing in our population may explain the high rate of co-existing middle ear fluid. There is no evidence to suggest an increased risk of SNHL among the paediatric ASD population.[7]

ABR is a very resource-intensive modality of hearing assessment. It is traumatizing, requires premedication, and may even have detrimental effects on a child's cognitive development. We suggest that there is a great need for developments in the area of hearing assessment in the clinic for children with ASD.

There are many limitations of our study. Due to the small sample size N=57, it is not possible to extrapolate our data to infer the incidence of hearing loss in the general ASD population. However, we have demonstrated the low diagnostic threshold of ABR in this population and the need for the development of less invasive, kinder assessment modalities which would ideally be based on in-office assessment.

In conclusion, our study demonstrates the low diagnostic yield of ABR under GA in the Irish paediatric ASD population. Given the known detrimental neurocognitive effects on the developing brain, the often-traumatizing experience of general anaesthetic in children with ASD, and the significant resource allocation involved with this procedure, we suggest that there is an urgent need for the development of reliable, objective, in-clinic hearing assessments modalities tailored to the ASD population.

Declarations of Conflicts of Interest:

None declared.

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21st March, 2024

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