Adel Zeglam¹ and Samah Al-Ksaik¹

¹Tripoli University Hospital, Tripoli, Libya. (Correspondence to: A. Zeglam: adelzeglam@gmail.com)

Abstract

Background: In low and middle-income countries where the health care is in a weakened state, spending money on unaffordable and probably unnecessary investigations might be substituted by a reliable, simple and more informative tool that can deal with the problem.

Aims: To examine current medical practice of measuring auditory brainstem response for all children with autistic spectrum disorder, and assess the value of this test in these children and its applicability in low and middle-income countries such as Libya.

Methods: We reviewed the medical records of all children with autistic spectrum disorder who presented to neurodevelopment clinics of Al-Khadra Teaching Hospital, Tripoli, Libya between January 2010 and December 2014.

Results: In 71 of 2368 children with autistic spectrum disorder, the family were concerned about their children’s hearing and reaction to loud noises. Auditory brainstem response confirmed that 26 of these 71 children had sensorineural hearing loss.

Conclusion: We consider auditory brainstem response measurement to be unnecessary in children with autistic spectrum disorder without clinical signs suggesting hearing impairment and without any parental concerns about hearing.
Introduction

Mental illness is the leading cause of disability worldwide, according to the World Health Organization. Most low- and middle-income countries including Africa have only 1 child psychiatrist for every 1–4 million people and this probably applies also to neurodevelopmental pediatricians and neurologists (1).

As stated in the report of the Commission on Social Determinants of Health (2): “Poverty is not only lack of income. The implication, both of the social gradient in health and the poor health of the poorest of the poor, is that health inequity is caused by the unequal distribution of income, goods, and services and of the consequent chance of leading a flourishing life. This is not in any sense a ‘natural’ phenomenon.”

The assessment and management of children with autistic spectrum disorder (ASD) is governed by codes of practice. The need for new recommendations is re-examined at regular intervals in the medical literature. However, despite the prevalence of ASD, agreement over management remains disputed; in particular, the need for hearing tests. According to the fifth edition of Diagnostic and Statistical Manual of the American Psychiatric Association (DSM-5) (3), ASD is a lifelong developmental disability defined by diagnostic criteria that include deficits in social communication and interaction, and restricted, repetitive patterns of behaviour, interests or
A 2014 survey of parents by the government of the United States of America (USA) suggested that 1 in 45 children, aged 3–17 years had been diagnosed with ASD (4). This is notably higher than the official government estimate of 1 in 68 American children with autism, as stated by National Centre for Health Statistics.

In Libya, a prospective hospital-based study of all children referred to a neurodevelopment clinic between 2005 and 2009 for ASD assessment showed a prevalence of 1 in 300 (5). In another study that was carried out in 2011, 15 085 children were seen and 200 aged 2–13 years were referred for ASD assessment. ASD were diagnosed in 83% (166 children) which gave a prevalence of 10:1000 (6, 7). The Libyan health system has been adversely affected since the beginning of the 2011 conflict. Many factors impose serious challenges to the public health sector in Libya such as the insufficiency of health information systems, severe shortage of medical supplies, and loss of health staff, which are mainly caused by the lack of security and interrupted delivery of supplies (8).

The prevalence of ASD in Libya is probably higher or similar to that seen in the USA and United Kingdom of Great Britain and Northern Ireland (UK). No data were available from the Arab countries or other developing countries. The increase in the estimated number of children with autism does not include children who are deaf. According to the Annual Survey of Deaf and Hard of Hearing Children and Youth that was conducted in the USA by the Gallaudet Research Institute in 2009–2010, 1 in 59 children (specifically those aged 8 years) with hearing loss were also receiving services for autism (9). In 2012, Szymanski et al. (10) showed that the prevalence of ASD in the USA was considerably higher in children with hearing loss than the reported national estimates of 1 in 91 (11) and 1 in 110 (12) for hearing children. This is approximately twice what is currently believed to be the national prevalence rate of 1 in 50 (3).

On average, a child who is deaf will be diagnosed with autism later in life than a hearing child will be. In 1 study, deaf children were diagnosed an average of 1 year later (13). Researchers and educators speculate that the reasons behind delayed diagnosis are the difficulty in distinguishing the characteristics of deafness from those of autism, as well as limited resources for parents and educators guiding the identification of autism and deafness. Also, it is possible that deaf children are diagnosed with ASD later because there are few psychological tests that have been devised, or even include considerations for children who are deaf. In fact, there are no approved instruments for making a diagnosis of autism in a child who is deaf (14). Some suggest that children with autism have language difficulties because they struggle with converting auditory information to vocal utterances (15). Others have suggested that children
with autism have severe auditory processing deficits (16). Deaf children might be occupied by self-stimulating behaviour and play independently, but this does not mean that the children have ASD but simply that they cannot hear other children. Knowing if the child has hearing impairment is not always apparent. The fact that hearing loss often is not diagnosed until children are 2 or 3 years old – the same time that autism is often diagnosed – increases the difficulty of assessment. Distinctive features of autism may mimic deafness and vice versa. If children cannot hear, they do not respond when called by name, which is often an important item on the checklist for autism, but it is also typical of deafness. Additionally, if children cannot hear, social interaction with peers might be difficult, due to an inability to hear conversation and not because they have autism.

Healthcare systems across developing countries including Libya face a number of formidable challenges. Limited energy resources, inaccessibility to medical facilities, limited government funding, and high costs are placing tremendous strain on the healthcare systems in general and on services for children with special needs in particular. In view of all these difficulties, this study examined the current practice of hearing tests in children with ASD in Tripoli, Libya.

**Methods**

**Study design**

The study was conducted between 2010 and 2014. A total of 292,633 children attended paediatric clinics for different medical reasons during this period, a diagnosis of ASD was made in 2368 children. In 71 (50 boys and 21 girls) of these children with ASD, ABR confirmed that 26 (19 boys and 7 girls) had sensorineural hearing loss (SNHL). The Neurodevelopment Clinics of
Al-Khadra Hospital, Tripoli are dedicated clinics in a tertiary care university hospital and are designated as referral and diagnostic clinics for autism and other neurodevelopmental problems. The hospital is affiliated to the Faculty of Medicine, Tripoli University. There are 3 regular weekly clinics. These clinics are consultant led (AZ), in addition to having 1 registrar, 2 senior house officers, and nurses. The clinics serve Tripoli, its suburbs and other district hospitals. The clinics receive referrals from other regions in Libya and care for children with other neurodevelopmental problems, such as hypotonia and epilepsy. An average of 40 children aged 1 month to 16 years are seen in each session. About 70% of the children attend the clinics for assessment of autism and speech and language disorders.

The inclusion criteria were age 1–16 years; speech and language disorders; and diagnosis of ASD and/or SNHL. The exclusion criteria were: neurometabolic disorders; genetic or infectious disease; chromosomal abnormalities; neurological or neurodegenerative disorders; and major physical abnormalities and congenital malformation.

**Diagnosis of ASD and screening for hearing loss**

A range of assessment tools were used to diagnose ASD, including the Modified Checklist for Autism in Toddlers (M-CHAT) (20), Reviewed Autism Diagnostic Interview (ADI-R) (21) and DSM-4 and 5 (3), and as well as observations in various settings and discussions with other professionals.

Auditory brainstem response (ABR) and otoacoustic emission (OAE) tests were used to screen for hearing loss. OAEs are sounds given off by the inner ear when responding to a sound. If the child's hearing loss is > 25 dB, they do not produce OAEs. All ABRs were acquired with a GSI 37 audio-screener (Grason–Stadler, Eden Prairie, MN, USA), which is a revolutionary hearing screening device that combines the 2-in-1 technology of measurement of OAEs and ABRs. For infants, only 1 sound is presented, which is called the click. If a healthy result is obtained, then the child passes the ABR test. The OAE test measures the reaction that is produced by the inner ear or cochlea. If an emission is produced for sounds that are essential to speech comprehension, then the child passes the test.

All children underwent complete general, neurological and neurodevelopment assessment. The results of the general and neurological examinations were unremarkable but the results of the neurodevelopment assessment were variable and needed to be re-evaluated in a separate session.
For each child, the parent was asked to respond to a brief demographic and social questionnaire covering the child’s age, gender, birth order, number of siblings, whether there were 1 or 2 parents at home, and whether the child had received professional psychological help.

The families were asked 5 questions regarding their child’s response, or lack of response, to an auditory stimulus, such as their name, by turning their head to maintain eye contact upon hearing it. Do they respond in other ways? Do they change their facial expression or make an eye shift when they hear their name? Do they turn away? Do they respond verbally?

Due to high illiteracy rates among rural parents, questionnaires were approved by a speech and language therapist and were administered through interviews at the clinic. The interviewers were either the registrar or any of the junior doctors attending the clinic who were given instructions by the investigator (AZ) about how to ensure consistency and avoid bias in completing the questionnaires. Due to the high cost of the ABR test, the control results for 700 children (440 boys and 260 girls) were retrospectively selected from records of children referred to the audiology department with speech and language disorders only, without any neurological and neurodevelopmental disorders.

**Control group**

The following criteria were used to choose the control group: age matching with the ASD group; ABR results including all stages performed in ASD children; and details that the neurological and neurodevelopmental examinations were unremarkable had been documented and confirmed by a consultant. All information obtained was entered on specially designed forms and kept in the medical files department. The names of the patients in the control group were not recorded and they were given a code number and sticker.

**Ethical considerations**

Parental approval and consent for the study were obtained. All examinations of children with ASD were performed in the presence of their parents. No consent was required for the control group as only patient files were used.

**Statistical analysis**

All statistical analyses were conducted using the descriptive tests in SPSS 20 statistical package. The risk of SNHL in the absence of other clinical signs and parental concerns was small (Results)
A total of 292,633 children attended the paediatric outpatient clinics for different medical reasons between 2010 and 2014; among whom 2,368 were diagnosed with ASD, giving a prevalence of 8:1000 (0.8%). A total of 2,809 children had only learning difficulties but no ASD or hearing deficits.

Among 107 children with SNHL, 26 had ASD. Prevalence of SNHL among children with ASD was 1.1%, while the prevalence of SNHL among all children seen in the Neurodevelopmental Clinic was 0.036%.

Two things are immediately obvious from the data shown in Table 1. The first is the low incidence of reaction to loud noises (3%) in children with SNHL. The second is the high rate of concern regarding hearing among parents of children with SNHL; 92% compared with only 8% of parents who had no concerns regarding their child’s hearing. Ninety-seven percent of parents of children with ASD had no concerns regarding hearing, supported by the fact that 98% of those children reacted to sounds and noises. Almost all children with developmental delay reacted to loud noises and their families had no concerns regarding their hearing or behaviour. However, they were concerned about delayed speech and language, which was obviously part of the children’s developmental delay and learning difficulties. Only 18% of children with ASD and SNHL reacted to loud noises, and 84% of parents had concerns regarding hearing. All of these children underwent ABR testing at the time of diagnosis. Most of them had other associated features of ASD.

Extrapolation of these data shows that in 71 of 2,368 children with ASD, the families were concerned about their children’s hearing and reaction to loud noises. ABR testing confirmed that 26 of these 71 children had SNHL. In children with ASD, with no signs and symptoms of hearing impairment and with no parental concerns, routine ABR is unjustified. We showed that relying on parents’ observation and concerns was a sensitive (87%) and specific (99%) predictor of SNHL in this group of children, which saved money on unnecessary investigation when resources were limited.

Discussion

The results of the present study suggest that ABR measurement is unnecessary in children with ASD without clinical signs of hearing impairment and without any parental concerns about
hearing. Our study has shown that relying on parents’ observation and concerns was a sensitive (87%) and specific (99%) predictor of SNHL in this group of children, which saved money on unnecessary investigation when resources were limited. Routine ABR testing in children with autism is probably unjustified and places a burden on families, as well as causing pain and distress to children.

Only a few studies have investigated the incidence of SNHL and ASD. A dual diagnosis was made in 1–6% of children who were deaf or hard of hearing. Rosenhall and colleagues found that the incidence of SNHL among children with ASD was 1.6% unilateral, 7.9% mild to moderate and 3.5% profound (22). Other studies have found an incidence between 5.3% (23) and 1.7% (24).

Speech and language delays and hearing disorders such as SNHL, hypersensitivity to sound, and otitis media are a few examples of conditions associated with autism (25). The management of children with ASD has changed dramatically over the past 15 years. However, there is still wide variation in practice between clinics and individual neurodevelopment paediatricians in the management of this common yet complex disorder. More than 70% of children in whom ASD is suspected would have undergone unnecessary hearing assessment because of a perceived risk of occult SNHL in this group of children. The available data from our study suggest that this is not justified. The risk of SNHL in the absence of other clinical signs and parental concerns is small.

Our study had some limitations. We did not collect information from other audiology services in other regions, which meant that we had no data on interventions provided at these centres. As none of these centres has ABR/auditory steady-state response equipment, we can only assume that children would be diagnosed by subjective means, which would usually take place at an older age. We feel that including data on these children would add weight to our study. We also did not collect data on the interval between diagnosis and initiation of interventions.

There is a need to develop an affordable and reliable tool for hearing assessment in resource-constrained, medically underserved areas of developing countries. There are many obstacles to the supply of proper health services in developing countries, including corruption in the supply of medical and therapeutic products, inconsistency in pricing, and wide fluctuation in quality and safety. Many children have difficulties in accessing essential health care because it is expensive and unaffordable. This situation is compounded by an absence of social security to ensure that poor people get the services they require, poor financial management, and lack of good administration and management. The current conflict in Libya has created new health needs. One critical problem is the lack of primary healthcare facilities, such as local clinics and district hospitals. Violence and general insecurity are making it difficult for children in Libya to reach health facilities and obtain the treatment they need. The lack of qualified personnel is
aggravated by the departure of foreign health workers, who used to make up a majority of health staff in the country. There are a lot of important areas that we need to address now, for example, mental health, autism and psychosocial support.

Our suggested practice after diagnosis of ASD in children in whom hearing impairment is suspected follows a “less is more” attitude. Infants are assessed according to local protocols and those without clinical signs suggesting hearing impairment and without any parental concerns are given open appointments. If no concerns are expressed and the children appear well, ABR testing is considered unnecessary. This approach might be applicable to all developing countries where the delivery of effective public health interventions to people in need is compromised and clinics struggle to offer services to local populations, particularly in remote rural areas.

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Test d’audition dans le trouble du spectre de l’autisme : est-il superflu dans les pays à revenu faible et intermédiaire ?

Résumé

Contexte : Dans les pays à revenu faible et intermédiaire où le système de soins de santé est fragilisé, le recours à des tests trop coûteux et probablement superflus pourrait être remplacé par un outil fiable, simple et plus instructif, permettant de s’attaquer au problème.

Objectifs : Examiner la pratique médicale en vigueur, qui consiste à mesurer les potentiels évoqués auditifs du tronc cérébral chez tous les enfants atteints d’un trouble du spectre de l’autisme (TSA), et évaluer la pertinence de ce test pour eux, ainsi que son applicabilité dans les pays à revenu faible et intermédiaire comme la Libye.

Méthodes : Nous avons passé en revue le dossier médical de tous les enfants atteints d’un TSA qui avaient consulté dans les cliniques de neurodéveloppement de l’Hôpital universitaire Al-Khadra à Tripoli (Libye) entre janvier 2010 et décembre 2014.
Résultats : Pour 71 des 2 368 enfants atteints d'un TSA, la famille s'inquiétait de leur audition et de leurs réactions aux bruits forts. Les potentiels évoqués auditifs du tronc cérébral ont confirmé que 26 de ces 71 enfants souffraient d'une surdité neurosensorielle.

Conclusions : Nous estimons que les potentiels évoqués auditifs du tronc cérébral sont superflus chez les enfants atteints d'un TSA sans signes cliniques évocateurs d'une déficience auditive et en l'absence d'inquiétudes de la part des parents concernant leur audition.
WHO EMRO | Hearing testing in autistic spectrum disorder: is it unnecessary in low and middle-income countries?

References


