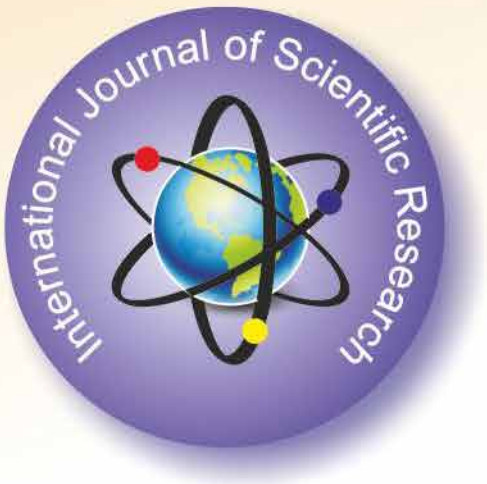
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Research Paper

Transient Auditory Dysynchrony Due to Non-Maturational Causes Evidenced by ABR – A Case Report

Medical Science

KEYWORDS : Dyssynchrony, recovery pattern, Auditory Brainstem Response

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ABSTRACT

Auditory neuropathySpectrum Disorder (ANSD) is a disorder which affects synchronous firing of auditory nerve and itsmanifested symptoms vary from individual to individual. A proportion of this population mainly infants below one year show spontaneous recovery which is attributed to maturation related factors. Presented here are two enigmatic case reports, one adult and one child,showingtransient ANSD and an interesting pattern of recovery of ABR, different from the traditional patternreported. Correlations with different tests and implications for further research have been discussed.

Introduction

Auditory Neuropathy Spectrum Disorder (ANSD) is a disorder characterized with normal Oto Acoustic Emissions (OAE) and/ or Cochlear Microphonics (CM) but absent or severely abnormal Auditory Brainstem Response (ABR) (Starr, Tremblay, Pasternak, & Kraus 1996). Its incidence among normal hearing individuals is reported to be 0.2- 0.3 % and 11-12% in the hearing impaired population (Ranceet al., 1999). Generally there is no relation between hearing thresholds and speech understanding and subgroup of ANSD reported to have fluctuant/ transient presence of ANSD has been reported by Berlin, Bordelon, Hurley, Hood, and Parkin (1997) in 5 infantsand Madden et al. (2002) in 9children. As an evidence for recovery, ABR has been shown to reappear, both partial (Attias & Raveh, 2007) and complete (Attias & Raveh, 2007; Berlin, 2003; Madden, Rutter, Hilbert, Greinwald, & Choo, 2002) and the major cause for this recovery is said to be maturational as evidenced by pattern of ABR recovery.

Participants and Methods

Case reports:

Case I:

A 39-year old female came with the complaint of on and off headache on left side associated with periorbital pain since 3-4 months, vomiting, and reduced hearing in left ear. On examination, motor & sensory systems, reflexes, visual acuity and visual field were found to be normal. However, cerebellar signs were present. Imaging reports were suggestive of multiple ring enhancing lesions in brain with the largest one in left occipital lobe with features of raised intra cranial tension. Hence, the client underwent left parieto-occipital space occupying lesion excision with duraplasty under General Anesthesia in lateral position. The final diagnosis made was Metastatic (brain) papillary adenocarcinoma with unknown primary. After the surgery, palliative radiation therapy and chemotherapy was planned.

Case II:

One year old female primi born in term to second degree consanguineous parents came with the complaint of delayed developmental milestones. On pediatric evaluations and laboratory investigation, the child was diagnosed to have microcephaly, spastic quadriplegia and increased lactate levels. In terms of speech and language, cooing was achieved by 3 months, babbling by 5 months and monosyllables by 9 months of age. The child recognizes family members and stranger anxiety was present.

Audiological Evaluation

Behavioral Tests

Case I: Prior to the administration of chemotherapy, a baseline audiogram was obtained. The client was concerned of a reduction in hearing ability in both the ears (left > right), with onset in right ear after surgery. She also complained of difficulty in comprehending speech (left > right), difficulty in localization, referred ear pain around pinna and tinnitus in left ear. Pure Tone Audiometry was done using GSI-61 Clinical Audiometer with TDH-50P headphones and B-71 bone vibrator for both right and left ear. Modified Hughson and Westlake method was used to estimate hearing thresholds.

Case II: Behavior Observation Audiometry (BOA) was done using the same instrument and set up as described above along with sound field system and ascending method was used to find out the lowest level at which the change in baseline behavior occurs. Two clinicians (tester and observer) had to agree for correct responses to be considered as a reliable threshold. Warble tone from 25Hz to 4000Hz and live speech served as stimulus.

Physiological and electrophysiological tests

Duly calibrated middle ear analyzer (GSI-Tympstar) was used for assessment of middle ear. Type 'A' tympanogram was observed in both the participants (compliance between0.3 & 1.75ml and middle ear pressure between +/- 100 dapa). ABR was recorded in both the subjects following natural sleep in an acoustically and electrically shielded room. Informed consent was obtained from the parents prior to administration of test procedures. Electrode sites were cleaned using NuPrep skin preparation gel and electrodes were placed on sites with Ten20 conductive gel. Absolute and inter electrode impedance were within 2 kOhms, and the electrode montage used was Fz-A2and Ethmotic research ER-3 insert earphones were used for stimulus presentation. Obtained EEG was online filtered between 100Hz and 3000Hz with artifact rejection of 23.8 micro volts and was epoched between -12 msec and + 12 msec. A total 1000 of stimuli were presented at a rate of 11.1/sec with alternating polarity and recording was repeated 3 times at each intensity constituting three averaged waveforms of 1000 stimuli. All the subjects were stimulated monaurally in both ears. ABR was recorded using IHS Smart EP Software, Version: 3.94 and BioLogic Navigator Pro.

Otoacoustic Emissions:

Transient Evoked Otoacoustic Emissions (TEOAE) was recorded in both the ears using ILO292 DP Echo port with V6 software. An SNR of 6dB and above was considered as presence of OAE.

Results

Initial Evaluation:

Case I:

Pure tone audiometry results revealed moderate sloping sensorineural hearing loss in the right ear and profound hearing loss in the left ear (Figure 1) with Speech Identification Scores (SIS) of 20% in right ear. TEOAE were obtained with good amplitude and reproducibility in both ears. ABR results showed no identifiable peaks and also the waveforms were non-replicable bilaterally (Figure 1). Considering the clinical findings and patient's complaint of inability to understand speech, a diagnostic label of auditory dysynchrony was made.

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Case II:
BOA response was observed as cessation of activity, smiling and rudimentary head turn consistently between 50 to 60 dBHL for warble tones from 250Hz to 4000Hz and for live speech. The OAE and ABR findings in case II is same as that of case I (Figure 1)

Follow up evaluation:
Case I
Patient returned after one month period on their own, reporting that there is a significant improvement in speech understanding. The same sequence of tests was repeated during this second evaluation. Pure tone audiometry results were no different from initial evaluation of moderate sloping sensorineural hearing loss in right ear and profound hearing loss in left ear. Speech audiometry results revealed a noticeable increment with speech identification scores of 75%. ABR was performed with the same protocol as before and results revealed replicable wave V at 80dBnHL. Even with reversal of polarity, rest of the waves is not identifiable (Figure 2)

Figure 2 about here

Case II:
Case was followed up after a period of one year, same test protocol was repeated and results were similar to that of case I described above. There were no changes in behavioral and OAE assessment results but ABR wave V was observed till 70dBnHL for click stimulus (Figure 2).

Discussion
Recovery in ANSD has been previously reported through both behavioral thresholds (Madden at al., 2002) and ABR thresholds (Attias & Raveh, 2007; Berlin et al., 2010). Behavioral thresholds on an average improved by 34.8dBHL in 6 months and ABR results showed both partial and complete recovery. The course of ABR recovery in the above said studies demonstrate complete absence of responses initially followed by appearance of wave I alone and wave III and V recovers progressively later. In contrast to these classical findings, the current study shows the recovery of ABR wave V initially with the absence of other earlier peaks. In literature, the above said description fits in to definition of "grossly abnormal ABR" where in which only wave V is prominent and replicable only at higher presentation levels (Berlin et al., 2010). From the current study it is worth to note that, the ABR recovered from complete absence to grossly abnormal ABR.

In literature, the sighting of grossly abnormal ABRs is not rare; however the reason for such a variation is less explained. To our knowledge the current study brings the first evidence to show atypical ABR recovery pattern in ANSD. This spontaneous recovery would be the possible reason for finding a "grossly abnormal ABR". Existing body of literature mainly attributes such spontaneous recovery in ANSD to be "maturational delay" and that is further evidenced by the fact that the studies included only infants. The above said explanation will not support the findings of the current study i.e., recovery seen in adult (case I) and recovery pattern didn't follow maturational process. Case I had Central Nervous System (CNS) disturbance which lead to loss of neural synchrony and recovery in speech understanding was seen following pharmacological and surgical intervention. Though the mechanism of these CNS disturbances affecting ABR and its recovery is unknown, this pattern of recovery (i.e., occurrence of wave V and not earlier waves) could suggest that, though the extent of loss of neural synchrony is common along the auditory pathway, its effect on lower centre is much greater than higher centre. This could be due the branching and overall increase in number of neurons as it goes higher in the auditory system and resulting in increasing neural synchrony sufficient to evoke wave V.

This may be consistent with adults who acquire ANSD at later ages and they demonstrate absence of ABR with consistently recordable Middle Latency Responses & Late Latency Responses. Another important observation seen in both cases is that there may not be any relation between observed behavioral thresholds and speech understanding ability during the time of recovery. With respect to the relation between wave V and speech understanding, ANSD is often viewed as a disorder which induces temporal disturbances and hence the loss of neural synchrony. Existing reports (Narne & Vanaja, 2008; Zeng, Oba, Garde, Sininger, & Starr, 1999; Zeng, Kong, Michalewsk, & Starr, 2005) suggest that there is a relation between extent of temporal disturbances and speech understanding. Contrary to existing views of ABR not being good indicator of extent of neural dyssynchrony (Madden et al., 2002), the current study showed that, there is a support for a positive correlation between ABR and speech understanding. Though the behavioral thresholds remained the same between two evaluations, improvement in speech understanding was consistent with recovery of wave V. Few studies supporting these findings are Abbas and Brown(1991a); Herman and Thornton(1992) and Gallégo,Truy, Morgon, and Collet(1997a). This non maturational dependent recovery of ANSD could even be found in children as in case II. These findings recommends audiologist for continual monitoring of auditory system and appropriate management options in this population.

Figures:

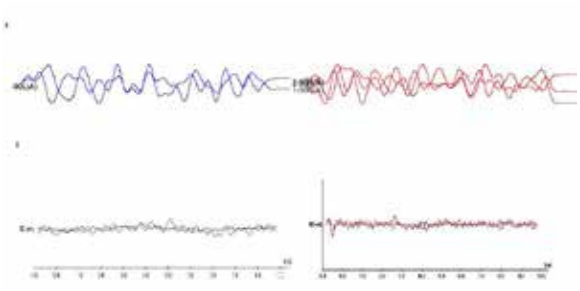


Figure 1: ABR for case I (B) and case II (A) at the time of first evaluation.

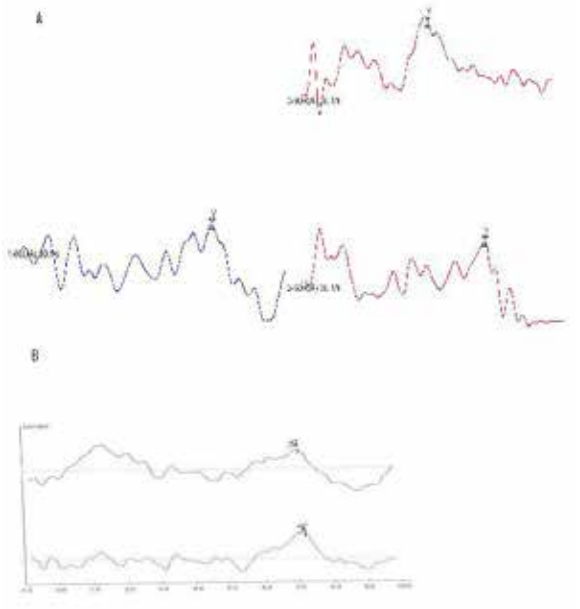
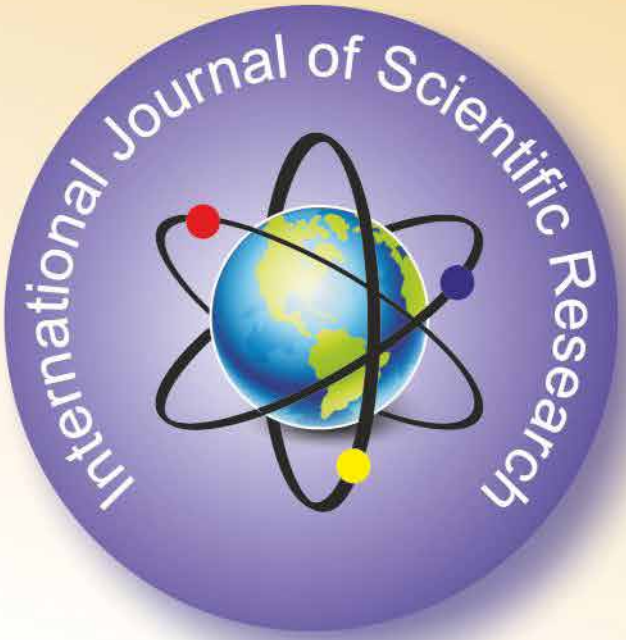


Figure 2: ABR for case I (B) and case II (A) at the time of follow up evaluation. Follow up period for case I is one month and case II is one year respectively.

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