Paper1

by Unnamed Author

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ABSTRACT

Objectives: Auditory brainstem implantation (ABI) can provide auditory stimulation in cases where cochlear implantation is contraindicated. The purpose of this study is to formally and informally assess the auditory, speech, and language development of Indian paediatric ABI users at regular post-operative intervals.

Design: Between January 2009 and April 2012, 5 children (13-94 months old) received an auditory brainstem implant. The children's auditory perception, speech intelligibility, and receptive expressive language development were accessed formally and informally at regular intervals up to 36 months device experience. All children attended post-operative habilitation sessions.

Results: All subjects' increased their auditory perception, speech intelligibility, and receptive language scores over time, although none achieved maximum scores on any test. Only 3 subjects were assessed beyond the 12-month interval. Development stagnated after the habilitation program ended. Informal assessment (AuSpLan) gave a more detailed and nuanced pictures of subjects' development.

Conclusion: Auditory brainstem implantation allowed paediatric users access to auditory stimuli and all subjects' development benefitted from the implantation. Informal assessment provided a more nuanced and complete picture of development than formal tests alone, and could be a valuable addition to test batteries. Clinics should consider providing equal emphasis on auditory and auditory visual based training to improve speech perception and success of communication; extending the post-operative habilitation support for longer duration and/or; developing an effective home-training program to maximize benefit from an auditory brainstem implant.

Keywords: Auditory Brainstem implant, AuSpLan,

Conflicts of Interest:

The authors report no conflicts of interest.

Introduction

The auditory brainstem implant (ABI) was designed for people with hearing loss due to severe inner ear malformations, complete cochlear ossification, or absence or non-functional auditory nerve (or Neurofibromatosis Type II) who would not benefit from a cochlear implant. The ABI bypasses the cochlea and auditory nerve and provides its users with an opportunity to detect and recognize auditory information through electrical stimulation of the auditory neurons of the cochlear nucleus.

Although adult ABI recipients do not attain the same levels of audiological ability as cochlear implant recipients (Schwartz et al. 2003, Sennaroglu et al. 2012), they are likely to benefit from improved lip reading ability (Lenarz et al., 2001; Schwartz et al., 2003; Behr et al., 2006; Maini et al., 2009) and improved speech perception, although the latter varies from limited (Lenarz et al., 2001; Nevison et al., 2002; Schwartz et al., 2003) to more substantial understanding (Jackson et al., 2002, Skarzynski et al., 2000, 2003; Bahr et al., 2006; Grayeli et al., 2008), especially in subjects without a tumour (Colletti V et al., 2005a, 2009). Some ABI users have been able to use the telephone (Lenarz et al., 2001; Sanna et al., 2006) although this is not an expected outcome.

The effects of ABI on children have been somewhat less broadly studied. Most published research comes from a single centre (Colletti V, et al., 2002, 2005b; Colletti L, 2007; Colletti L et al. 2008), who has shown that with an ABI (and presumably associated regular habilitation), children, even those with additional needs, often achieve good to moderate speech detection and occasionally open-set speech recognition. These finding have been echoed by Choi et al. (2011), Sennaroglu et al. (2009), and Goffi-Gomez et al. (2012).

Paediatric ABI is gaining popularity in India. Due to its sheer size, India has a massive number of potential ABI candidates, thus there is an important and growing need to document and assess the speech perception and language development outcomes of children who have received an ABI and the post-implantation habilitation they need to attain maximum benefit.

Methods and Materials

Subjects

Inclusion criteria for subjects was as follows: to have been implanted with an ABI at the Madras ENT Research Foundation clinic between January 2009 and April 2012, to have received a MED-EL (Innsbruck, Austria) ABI, and to be willing and able to commit to the habilitation program.

Pre-operative Protocol

High resolution computerized tomographic (HRCT) scans and magnetic resonance imaging (MRI) of the temporal bone were used for pre-operative diagnosis. Subjects hearing status was evaluated by both objective and subjective audiologic measurements (pure tone audiometry, immitance, acoustic reflex, auditory brainstem response, otoacoustic emission (OAE) and auditory steady state response).

Surgical Approach and Post operative evaluations

The auditory brainstem implant was placed in the brainstem, through the retromastoid craniotomy approach. During discharge, post surgery, the subjects were conscious, oriented, with no spino motor deficits and with hearing unchanged from the pre-operative status. The activation of audio processors took place within 3 months post operatively, in an intensive care unit with close and continous monitoring of the subjects' vital functions. The mapping audiologist provided maps with appropriate current levels that contributed to improved speech perceotion but did not elicit non auditory responses. Follow up mapping sessions took at the following post-operative intervals: 1 month, 3 months, 6 months, 1 year, 1.5 years, 2 years, and 3 years after initial ABI activation.

Assessment: Tests

The test battery for formal assessment consisted of the Categories of Auditory Performance (CAP) (Archbold et al. 1995) (see Table 5), Speech Intelligibility Rating (SIR) (Allen et al. 1998) (see Table 6), and Receptive Expressive Emergent Language Scale (REELS) (Bzoch et al. 1991) to assess subjects' auditory perception, speech production, and receptive and expressive language skills.

For informal assessment, Auditory, Speech, and Language (AuSpLan) Pyramids (McClatchie & Therres 2003) were used to assess subjects' auditory perception, speech production, and expressive language skills. AuSpLan, acronym of Auditory, Speech and Language, is a developmental curriculum for children with hearing impairment to learn to listen and develop verbal language. It details hierarchies of skills in three domains – Audition, Speech and Language – represented in the form of pyramids, which were used in the present study for assessment purpose. AuSpLan helps professionals categorize pediatric cochlear implant (CI) recipients into three groups (Commensurate, Capable and Challenged) based on factors that influence outcomes with a CI. Further it provides timelines for specific skill levels that each group of CI recipient can be expected to achieve.

Assessment: Intervals

The assessments took place pre-operatively and then at the following post-operative intervals: 1 month, 3 months, 6 months, 9 months, 1 year, 2 years, and 3 years after first-fitting. Results were cross-verified with each subject's lesson plans, progress reports, and video analyses of rehabilitation sessions during discussion meetings with the relevant rehabilitation professional.

Habilitation Program

All subjects attended a habilitation program at MERF-Institute of Speech and Hearing (a Unit of Madras ENT Research Foundation), Chennai. A developmental approach to communication and language acquisition was opted for all the subjects. The habilitation program consisted of twice a week sessions for the first 12 months of device experience and, thereafter, a follow-up visit every 6 months. Every session included goals for audition, language, cognition, speech and natural conversation during play. Summary of the session and home training tips were provided to parents in end of the session. Undue emphasize on both i) avoiding visual cues (lip cues) always by covering the speaker's mouth or talking from behind the back of the subject and ii) providing visual cues (lip cues) during conversation through exaggerated articulatory movements and very loud speeech were discouraged in the program

Results

Subjects

5 profoundly deaf children (mean age = 4y11m at implantation; 3 males and 2 females) met the inclusion criteria and were included in the study. 4 had Michel's aplasia, 1 (subject 4) had an absent auditory nerve. All subjects received a MED-EL (Innsbruck, Austria) PULSAR ABI with an OPUS^2 audio processor. 3 subjects (#1, 2, and 3) completed their 3 years of

implant use whereas 2 subjects (#4 and 5) had their implants only for 12 months (see Table 1). Subjects 1 and 2 each had an active channel turned off between first and last fitting so as to avoid possible non-auditory stimulation. Because subject 4 experienced facial muscle twitching at higher current levels, the maximum comfortable loudness levels were kept lower and 3 electrodes were turned off at first-fitting, and an additional electrode at last fitting. No additional adverse events were observed.

Subjects' actual participation in the habilitation program varied according to the parents' willingness to participate, their distance from the therapy centre, and economic status. The subjects' results were not statistically analysed due to their small number and demographic heterogeneity. Results were instead represented in graph form, as this allows a more meaningful interpretation.

Test results

1. Auditory results: Categories of Auditory Perception (CAP) and AuSpLan

All subjects scored 0 pre-operatively, and 4 or 5 at the 9-month interval on the CAP test (See Figure 1). After the 9-month interval only 1 subject (#1) improved his/her score. No subject at any interval scored the test maximum (7 points). The AuSpLan Auditory Pyramid shows a more detailed and nuanced picture of qualitative progress throughout the test period (see Table 2).

2. Speech results: Speech Intelligibility Rating (SIR) and AuSpLan

All subjects' SIR scores improved over time. A definite pattern of improvement, however, couldn't be observed (see Figure 2). When subjects' speech intelligibility development was tracked with the AuSpLan pyramid, subjects showed a steady qualitative improvement over time (see Table 3). Subject 5 had ‘word level intelligibility’ pre-operatively, while others were at pre-speech level. Subject 5’s progress is shown in grey so that it does not confound the trend exhibited by the other subjects.

3. Language skills results: Receptive Expressive Emergent Language Scales (REELS) and AuSpLan

ABI use facilitated the development of all subjects’ receptive and expressive language (see Figures 3 and 4). Not surprisingly, subjects tended to have higher receptive language scores than expressive language scores at the same intervals.

4. Informal assessment: AuSpLan

Assessed informally with the AuSpLan – Expressive Language Pyramid, all subjects developed their expressive language skills over time (see Table 4). As subject 5 had a higher pre-operative language score than the others subjects, /her scores are in grey. The improving trend seen in REELS score was also seen in the informal assessment.

Discussion

ABI is, by now, a standard treatment method for providing auditory stimulation to children who cannot benefit from cochlear implants due to inner ear malformations or auditory nerve damage. Expectations for auditory and speech development have increased from ABI being an aid for lip-reading to now enabling some users to develop open-set speech perception and intelligible speech (Otto et al. 2002), although these results are not typical (Schwartz et al. 2008, Merkus et al. 2013). That the success of these results is predicated on 1) subjects’ regular attendance and participation in habilitation programs and 2) at home support and training they receive from their parents/guardians (Schwartz et al. 2008) has perhaps been underemphasized in the literature, and, as such, it is all the more important to emphasize that for a child with prelingual hearing loss, learning how to first differentiate the apparent chaos of speech from environmental noise and then transform it into intelligible linguistic input is a skill that also needs to be practiced outside the clinic. Training parents to be encouraging and interactive with their child is especially important.

On the formal tests (CAP, SIR), subjects tended to develop steadily up to the 9^{th} or 12^{th} month interval, whereupon development slowed or stagnated. On the CAP, for example, none of the 3 tested subjects increased their score after the 12^{th} month interval and on the SIR test, only 1 of the 3 tested subjects increased their score after the 12^{th} month interval. Subject 2's SIR score increased from a pre-operative '1' to a '3' at 12 months, and was still a '3' at the 36^{th} month interval. And Subject 3's CAP and SIR scores didn't increase at all after the 9^{th} month interval, likely due to his/her receiving very poor home training and discontinuing habilitation after only 5 months of device experience. Looking at the CAP and SIR results, especially post-12^{th} month interval, would seem to suggest that the subjects simply stopped developing. This, however, is not entirely true. Initially, we found it difficult to believe that subject #s 1, 2, and 5 reached scores of 5 after only 9-12 months device experience. However, upon further investigation, we found that each of these subjects had either high preoperative language skills (subject #5) or very supportive parents/mothers who regularly attended habilitation sessions with them (subjects #1 and #2). Subject #2 in particular had, as the clinicians who worked with her repeatedly reported, "wonderful parents". Though not conclusive, supportive family environment appears to be a factor that positively influences outcomes with ABI.

Subjects exhibited steady development when their auditory development and speech production abilities were assessed (informally) with the AuSpLan (see Tables 2-3). The reason for this is that the abilities necessary to ascend the AuSpLan's hierarchy of skills are easier than those in the CAP and SIR; for example, on the CAP, a score of '6' ("Understands conversation without lip-reading with a familiar talker") or a '7' (Can use the telephone with a familiar talker) are very challenging to a paediatric ABI user. The goal on the AuSpLan auditory pyramid: "Comprehension of simple questions", is more attainable. The AuSpLan can document the children's incremental developmental progress in more detail that the formal tests; thus it could help hearing professionals show parents that progress is indeed being made. For this reason, we believe the AuSpLan pyramids and similar informal tools should be used in conjunction with the usual formal tests to evaluate developmental progress.

Regarding the subjects' receptive language age, all subjects except Subject 3 showed steady development over the course of the study, to the extent that their receptive language age was equal to or exceeded their length of device use. As was mentioned earlier, Subject 3 had very poor home training and discontinued habilitation sessions at 5 months, and which could be a reason that his/her receptive language age did not increase after 6 months device experience. Subject 5 enjoyed higher scores than the other subjects at every interval in which she was tested because she 20-74 months older than the other subjects at time of implantation, and thus more cognitively mature.

When evaluated by the REELS, the expressive language ages of all subjects, other than Subject 5, increased for 0-3 months before implantation to at least 12-18 months at the 12^{th} month interval. Then, stagnation set it: only 1 subject (Subject 1) appeared to mature between the 12^{th} and 36^{th} month intervals. In other words, up to the 12^{th} month interval, all subjects' expressive language age matched their length of device use; by the 36^{th} month interval, 2 of the 3 subjects tested were at least a year behind and 1 (Subject 2) was age equivalent. When evaluated with the AuSpLan hierarchy, subjects did exhibit steady growth. By the 24th month interval, all subjects were capable of "connected utterances at phrase level".

All the subjects' receptive and expressive skills benefited from receiving an ABI. Their active qualitative progress was particularly evident in informal testing with the AuSpLan pyramids. Reflections from informal assessments can be directly applied to plan future goals and help in parent counselling.

Considering that pediatric ABI recipients benefit from auditory-visual (lip reading) approach to communication and language learning there is a trend amongst professionals to focus loosely on auditory skill development. The subjects of the study showed improvement in their listening abilities with structured auditory training. It must be understood that a habilitation program for these recipients should lay simultaneous emphasis on both i) bottom-up (structured) auditory training for development of listening abilities, from simple to complex levels and ii) top-down (connected speech) auditory-visual based training for improving speech perception at conversational levels.

India is to some extent plagued by a lack of commitment to sustained habilitation: only 45.5% of the cochlear implant clinics in India counselled the parents of children with a CI to commit to a rehabilitation program for more than 1 year and only 58% of cochlear implants users continued habilitation after 1 year (Jeyaraman 2012). The results of the study showed that outcomes with an ABI tend to improve over years after implantation, indicating that ABI recipients would derive greater benefit with long term support, extending beyond one year, post implantation. Habilitation programs for pediatric ABI recipients should consider taking steps to foster long-term parent-clinic contact and encourage regular visits, and training parents to be supportive by giving them home training tips. Tele-therapy services and creation of satellite habilitation units could also be considered. Future studies, with a larger more homogenous subject population are needed to confirm the utility of informal assessment like AuSpLan as an addition to formal testing.

Conclusion

Children with an ABI develop audition, speech, and language skills gradually. Formal test outcomes in this study were encouraging but were restricted. Informal assessment allowed a more detailed picture to emerge and thus can be useful for clinicians and parents. The importance of attending a regular habilitation program and quality at-home verbal interaction with parents for children's development should not be underestimated.

Acknowledgments

We would like to thank the pediatric ABI users and their families for their support and cooperation.

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Figure Legends

Figure 1

Figure 1: Categories of auditory perception (CAP) scores over time

Figure 2

Figure 2: Speech intelligibility Rating (SIR) scores over time

Figure 3

Figure 3: Receptive Language Age (RLA) scores over time

Figure 4

Figure 4 : Expressive Language Age (ELA) scores over time

Table Legends

Table 1

Table 1: Demographic data of paediatric auditory brainstem implant (ABI) users

Table 2

Table 2: Timeline for development of auditory skills in paediatric ABI users using AuSpLan – auditory pyramid. Horizontally shaded squares represent the time when all subjects had achieved the hierarchy task.

Table 3

Table 3: Timelines for development of speech intelligibility in paediatric ABI users using AuSpLan – speech intelligibility pyramid. Horizontal shaded squares represent the time when all subjects had achieved the hierarchy task. Note: CVCV= consonant, vowel, consonant, vowel.

Table 4

Table 4: Timelines for development of expressive language skills in paediatric ABI users. Horizontal shaded squares represent when all subjects had achieved the hierarchy task.

Table 5

Table 5: Categories of Auditory Performance (CAP)

Table 6

Table 6: Speech Intelligibility Rating (SIR)

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