Research Article

Auditory, Speech, and Language Outcomes in Paediatric Auditory Brainstem Implant Users: an Indian Experience

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Key Words

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Abstract

Auditory brainstem implantation (ABI) can provide auditory stimulation in cases where cochlear implantation is contraindicated. The purpose of this study was to assess the development of auditory, speech, and language skills of Indian paediatric ABI recipients. Five children between January 2009 and April 2012, with ages ranging from 13 to 94 months received an auditory brainstem implant. The auditory, speech, and language development of the participants were assessed using formal and informal assessment tools, at regular intervals up to 36 months after activation of audio processor of the ABI. All these participants attended post-operative auditory habilitation sessions. There was an improvement in all the participants in terms of auditory perception, speech intelligibility, and, receptive and expressive language scores over time, although none achieved maximum scores on any test. Only three participants were assessed beyond the 12-month interval. The development stagnated after the habilitation program ended. Informal assessment (AuSpLan) gave a detailed information regarding development of the participants in these three domains. Auditory brainstem implantation provided access to sounds in environment and supported development of auditory, speech, and language skills in paediatric recipients. Informal assessment tools provided a more nuanced and complete picture of development than formal tests alone, and could be a valuable addition to the test batteries. The auditory habilitation professionals should consider the skills and needs of an ABI recipient, prior to choosing an appropriate communication approach for habilitation. Further aspects to be considered include 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.

Background

The auditory brainstem implant (ABI) was designed for people with hearing loss due to severe inner ear malformations, complete cochlear ossification, or absence / non-functional auditory nerve or Neurofibromatosis Type II who would not benefit from a cochlear implant (CI). 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 cochlear nucleus. The 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 in terms of improved speech 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 a 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 et al., 2005a, 2009). Some ABI users have even 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 et al., 2002, 2005b; Colletti , 2007; Colletti et al. 2008), which 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 findings have been echoed by Choi et al. (2011), Sennaroglu et al. (2009), and Goffi-Gomez et al. (2012).

In India, the number of paediatric ABI recipients has increased gradually over the last ten years. Auditory brainstem implantation in children is gaining popularity in India. There is an important and growing need to document the auditory, speech and language outcomes of children who have received an ABI. There is also a need to identify approaches and strategies to maximize outcomes of post implant auditory habilitation program.

Materials and Methods

Participant Inclusion Criteria

Children implanted with a MED-EL (Innsbruck, Austria) ABI implant system at Madras ENT Research Foundation clinic (Chennai, India) between January 2009 and April 2012 and enrolled for auditory habilitation program at the same clinic were considered for inclusion in the study.

Pre-operative protocol

The hearing status was evaluated by both subjective and objective audiologic measurements (pure-tone audiometry; immittance - tympanogram, acoustic reflex; otoacoustic emission (OAE), and auditory steady state response). The high resolution computerized tomographic (HRCT) scans and magnetic resonance imaging (MRI) of the temporal bone were used for pre-operative radiological evaluation.

Surgical approach and post operative evaluations

The internal part of the auditory brainstem implant was surgically implanted through the retromastoid craniotomy approach. After the surgery, during discharge, the participants were conscious, oriented, with no spino motor deficits and with hearing unchanged from the pre-operative status. The switch-on/ activation of audio processors took place within three months after implantation, in an intensive care unit with close and continuous monitoring of thevital functions of the participants. The audiologist provided maps with appropriate current levels that contributed to improved speech perception but did not elicit non-auditory responses. Follow-up mapping were performed at 1 month, 3 months, 6 months, 1 year, 1.5 years, 2 years, and 3 years after initial ABI activation.

Assessment

The test battery comprised of both formal and informal assessment tools.

a. Categories of auditory performance (CAP; Archbold, Lutman, & Marshall, 1995) is a rating scale consisting of eight performance categories, relating to auditory perception. It is arranged in a hierarchy of skills that increase in difficulty. The rating scale ranges from 0 to 7. A CAP of 0 indicates no awareness of environmental sounds (CAP 0); CAP of 1 indicates awareness of environmental sounds (CAP 1); CAP of 2 indicates response to speech sounds (CAP 2); CAP of 3 indicates recognition of environmental sounds (CAP 3); CAP of 4 indicates discrimination of at least two speech sounds (CAP 4); CAP of 5 indicates understanding ability of common phrases without lip reading (CAP 5); CAP of 6 indicates understanding ability of conversation without lip reading with a familiar talker (CAP 6), and a CAP of 7 indicates using the telephone with a familiar talker (CAP 7). It is widely used in paediatric cochlear implant research to monitor the auditory outcomes over time.

b. Speech Intelligibilty Rating (SIR; Allen Nikolopoulos, & O'Donoghue, 1998) is a simple and reliable clinical measure of speech intelligibility. This is a five-point rating scale with levels arranged in order of improving speech intelligibility. A SIR of 1 indicates pre-recognisable speech or manual communication (SIR 1); SIR of 2 indicates single word intelligibility (SIR 2); SIR of 3 indicates connected speech is intelligible to a listener who concentrates and lip reads within a known context (SIR 3); SIR of 4 indicates connected speech is intelligible to a listener who has little experience of a deaf person's speech (SIR 4); and SIR of 5 indicates connected speech that is intelligible to all listeners with the child being easily understood in everyday contexts.

c. Receptive Expressive Emergent Language Scales (REELS; Bzoch et al., 1991) is a norm referenced scale to assess receptive and expressive language abilities of children from birth to three years of age, through observational and caregiver interview method.

d. The Auditory Speech and Language (AuSpLan; McClatchie & Therres 2003); a manual for professionals working with children who have cochlear implants or amplification 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, i.e., Audition, Speech, and Language, represented in the form of pyramids, which was used as an informal assessment tool in the present study.

Assessment Intervals

The assessments took place pre-operatively and then at 1 month, 3 months, 6 months, 9 months, 1 year, 2 years, and 3 years after the first-fitting. The results were cross-verified with lesson plans,

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

Subject	Etiology	Age at implantation	Ear implanted	Hearing aid use (months)	ABI use (months)	Coding strategy	No. of active channels		Adverse events	Associated problems	Commitment to habilitation program
							First fitting	Last fitting			
1	Michel's aplasia	1y 11m	Right	6	51m	FSP	12	11	None	None	2yrs (follow up every 6m)
2	Michel's aplasia	4y 5m	Right	3	36m	HDCIS	12	11	None	None	1yr (follow up every 6m)
3	Michel's aplasia	6y 2m	Right	did not use	36m	FS4	12	12	None	None	Discontinued habilitation at 5m. Very poor home training
4	Absent auditory nerve	4y	Right	1	12m	HDCIS	9	8	Facial muscle twitching at higher current levels	refractive error - corrected with eyeglasses	1yr (continuing)
5	Michel's aplasia	7yrs 10m	Right	4	12m	HDCIS	12	12	None	None	1yr (continuing)

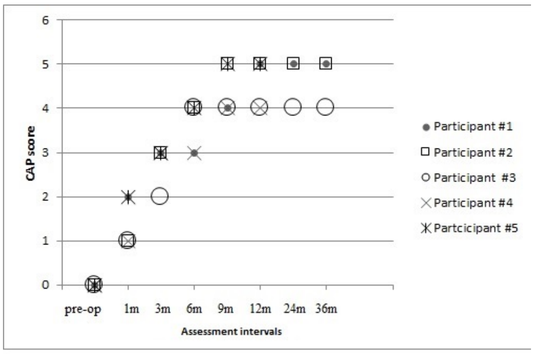

Figure 1: Categories of auditory perception (CAP) scores at pre- and post- implantation assessment intervals.

progress reports, video analyses of habilitation sessions and discussion with the relevant auditory habilitation professional of each participant.

Auditory Habilitation Program

All the participants were enrolled for auditory habilitation program at MERF CI speciality clinic, post implantation The program followed an 'auditory- oral/verbal approach to communication' with the recipients. In this approach, the development of listening and spoken language was promoted and undue emphasize on visual assistance (lip reading) to speech understanding was discouraged. The protocol comprised of sessions scheduled twice a week for the first 12 months post implantation and, thereafter, a follow-up visit every 6 months. Each session was of 60 minutes duration and included goals for listening, speech, language, and cognition, by engaging in natural conversation during play. A summary of the session and home training tips were provided to parents at the end of each session.

Results

Participants

Five profoundly deaf children (mean age = 4 years 11 months at implantation; 3 males and 2 females) met the inclusion criteria and were included in the study. Four had Michel's aplasia and one (subject 4) had an absent auditory nerve. All the participants received a MED-EL (Innsbruck, Austria) PULSAR ABI with an OPUS2 audio processor. Three participants (#1, 2, and 3) completed their three years of implant use whereas two participants (#4 and 5) had their implants only for 12 months. Participants 1 and 2 had an active channel turned off between first and last fitting so as to avoid possible non-auditory stimulation. As Participant 4 experienced facial muscle twitch-

Table 2: Time-line for development of auditory skills in paediatric ABI users using AuSpLan - Auditory Pyramid.

Auditory Hierarchy	Pre-op	1 m	3 m	6 m	9 m	12 m	24 m	36 m
No awareness	5							
Auditory awareness to speech		5						
Spontaneous awareness in distraction		2	2	1				
Suprasegmental discrimination of vocal length differences		1	3	1				
Suprasegmental discrimination of word length differences			1	4				
Segmental identification of words of same length			1	-	3			
Identification of one target in sentence context				1	-	2		
Identification of 3 targets in sentence context				1	-	-	1	-
Comprehension of simple questions					1	-	-	-

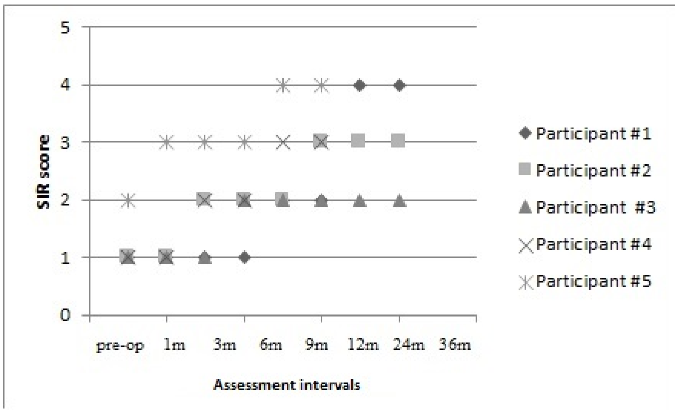

Figure 2: Speech Intelligibility Rating (SIR) scores at pre- and post- implantation assessment intervals.

ing at higher current levels, the maximum comfortable loudness levels were kept lower and three electrodes were turned off at first-fitting, and an additional electrode at last fitting. No additional adverse events were observed. The actual participation of the participants in the habilitation program varied according to the willingness of the parents to participate, the distance from the therapy centre, and economic status. The demographic details of the participants are mentioned in Table 1.

The data were not statistically analysed due to their small number and demographic heterogeneity. The results were instead represented in a graph format, as this allowed a more meaningful interpretation.

Auditory outcomes: Categories of Auditory Perception (CAP) and Auditory Pyramid of AuSpLan

All the participants scored 0 pre-operatively, and 4 or 5 at the 9-month interval on the CAP scale as in Figure 1. After an interval of 9 months, only 1 participant (#1) improved his/her score. None of the participants scored the test maximum score (i.e., 7 points) at any test interval. The development of auditory skills in the participants based on AuSpLan-Auditory Pyramid is given in Table 2. All but one participant (#3) achieved 'closed set auditory identification of words' and only one participant (#5) achieved 'auditory comprehension of simple questions' at the 9-month interval.

Speech outcomes: Speech Intelligibility Rating (SIR) and Speech Intelligibility Pyramid of AuSpLan

The SIR scores of all the participants improved over time. A definite pattern of improvement, however, could not be observed, as in Figure 2. When AuSpLan pyramid was used to track speech intelligibility skills of participants during post activation intervals, the pattern of improvement became clearer as seen in Table 3. Four participants (#1-4) achieved 'consonant-vowel sequences level intelligibility' at the 9-month interval. Beyond this in-

Table 3: Time-line for development of speech intelligibility skills in paediatric ABI users using AuSpLan - Speech Intelligibility Pyramid

Speech intelligibility Hierarchy	Pre-op	1m	3m	6m	9m	12m	24m	36m
Pre-speech behaviour	4							
3 basic vowels in isolation		2	2					
3-5 basic consonants in isolation				4				
Imitates CVCV sequences				1	3			
Spontaneously produces words with 80% intelligibility	1	-	-	-	1	-	1	1
Produces 2-3 word phrases with 80% intelligibility				1	1	-	-	-
Spontaneously produces 3-4 word phrases with 80% intelligibility					1	-	-	-

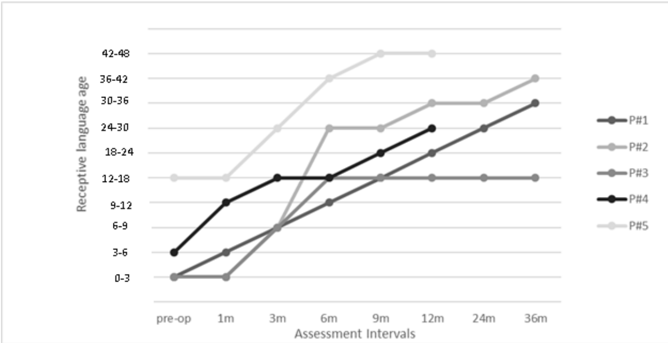

Figure 3: Receptive Expressive Emergent Language (REELS)- Receptive Language Age Scores at pre- and post-implantation assessment intervals.

telligibility level, the participants showed individual differences in achieving higher skills. One participant (# 5) had ‘word level intelligibility’ pre-operatively, while others were at pre-speech level. The progress made by this participant (#5) is shown in grey so that it does not confound the trend exhibited by the other participants.

Receptive and Expressive Language outcomes: Receptive Expressive Emergent Language Scales (REELS) and Expressive Language Pyramid of AuSpLan

The use of ABI facilitated the development of receptive and expressive language among all the participants, as depicted in Figure 3 and Table 4. Not surprisingly, the participants tended to have higher receptive language scores than expressive language scores at the tested intervals. The verbal expressive language skills of participants were assessed informally using the AuSpLan - Expressive Language Pyramid and the trend in improvement is showed in Table 4. All the participants achieved ‘word approximation abilities’ by the 9-month interval. Word and phrase level production were achieved. Only one participant (#5) achieved ‘basic sentence’ level spoken language abilities. As this participant (#5) had a higher pre-operative language score than the others participants, her scores are in a lighter shade of grey, in order not to confound with scores of other ABI recipients.

Discussion

The ABI is 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, The outcomes with an ABI are varied. Previously ABI, with its limited auditory benefits, was only viewed as an audiologic management option that provides additional cues to assist in speech reading; but now it is observed that an ABI can enable some users to even 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).

The auditory, speech, and language outcomes with an ABI may show individual variations based on several factors such as intrinsic (for example, status of cochlea and auditory nerve, cause of hearing loss and period of hearing deprivation, age at implantation, pre-operative listening and language skills, etc) and extrinsic (implant technology, com-

Table 4: Time-line for development of expressive language skills in paediatric ABI users using AuSpLan - Expressive Language Pyramid

Expressive language hierarchy	Pre-op	1m	3m	6m	9m	12m	24m	36m
Pre-speech behaviour	4							
Vocalization on imitation		3	1					
Spontaneous meaningful vocalizations			2	2				
Imitates word approximations (5-10 words)	1			3	1			
Spontaneous word productions (30-50 words)			1		1	1	2	
Connected utterances at phrase level				1		1	1	
Connected utterances at basic sentence level					1			

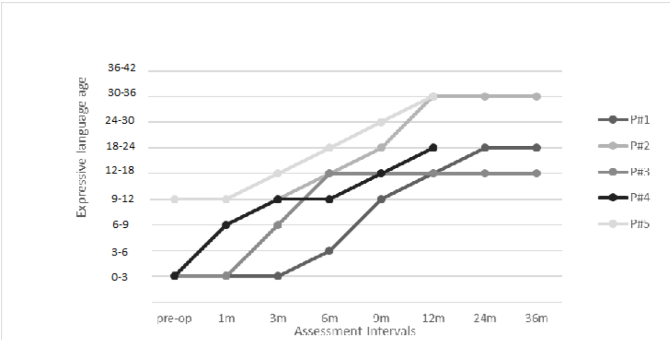

Figure 4: Receptive Expressive Emergent Language (REELS) - Expressive Language Age Scores at pre- and post- implantation assessment intervals.

mitment to post operative habilitation, quality of intervention services, home training and support, etc) factors. The importance of these extrinsic factors has perhaps been under emphasized in the literature (Schwartz et al., 2008).

On the formal tests (CAP, SIR), the participants tended to develop steadily up to the 9th or 12th month interval, and later the development slowed or stagnated. On the CAP, for example, none of the three participants increased their score after the 12th month interval and on the SIR test; only 1 of the 3 participants increased their score after the 12th month interval. The SIR score of one participant (#2)increased from a pre-operative '1' to a '3' at 12 months, and was still a '3' at the 36th month interval. and the CAP and SIR scores in on eof the participants (#3)did not increase at all after the 9th month interval, likely due to his/her receiving very poor home training and discontinuing auditory habilitation after 5 months of device experience. Looking at the CAP and SIR results, especially at 12th month interval, would seem to suggest that the participants simply stopped developing. This, however, is not entirely true. Initially, it was difficult to believe that three participants (1, 2, & 5) reached scores of 5 after only 9-12 months of experience with the device. However, upon further investigation, it was found that each of these participants had proper electrode placement in brainstem indicated by clear eABR peaks (Participant #5); high preoperative language skills (Participant #5), long-term commitment to rehabilitation (Participants # 1 & 2) or very supportive parents/mothers who provided training at home for listening and spoken language (participants 1 and 2). Participant 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 the outcomes with ABI.

All the participants exhibited steady progress in their auditory, language, and speech development when assessed with informal tool such as AuSpLan Pyramids in comparison to formal assessment. The reason for this is that the abilities necessary to ascend the hierarchy of skills in AuSpLanare 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) is very challenging to a pediatric ABI user. On other hand, AuSpLan being a curriculum that supports children with hearing impairment in developing listening and spoken language skills, the skill levels are broken down into smaller achievable units, that are placed in developmental order, from simple to more complex ones. The breakdown and detailing of each skill level of AuSpLan allows for monitoring the dynamics of development, over longer post implantation intervals, without getting stagnated at a level. For this reason, the AuSpLan pyramids and similar informal tools would be more useful and hence needs to be used in conjunction with the usual formal tools/tests to evaluate the developmental progress.

Regarding the receptive language age of the participants, all the participants (except # 3) showed a 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, one of the participants (# 3) had very poor home training and discontinued habilitation sessions after 5 months, and this could be a reason that his/her receptive language age did not increase after 6 months of experience with the device. One of the participants (# 5) attained scores higher than the others at every interval in which he/she was tested because he/she was 20-74 months older than the other participants at the time of implantation, and thus relatively more matured cognitively.

When evaluated by the REELS, the expressive language ages of all the participants, other than Participant # 5, increased for 0-3 months before implantation to at least 12-18 months at the 12th month interval. Then, stagnation in the development of skills set in. Only one participant # 1) appeared to mature between the 12th and 36th month intervals. In other words, up to the 12th month interval, the expressive language age matched the length of device use in all the participants. By the 36th month interval, 2 of the 3 participants tested were at least a year behind and one (Participant #2) was age equivalent. When evaluated with the AuSpLan Pyramid, participants did exhibit steady growth. By the 24th month interval, all participants were capable of 'connected utterances at phrase level'.

There was an improvement in the receptive and expressive skills of all the participants after 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 counseling.

As the outcomes are sometimes limited and varied with an ABI, habilitation professionals do realize that not all ABI recipients are candidates for an auditory and oral-verbal communication approach; that a few may need visual assistance or supports (speech reading) to assist in language learning or as a permanent choice of communication mode. For these reasons, there is a trend amongst professionals to focus lightly on auditory skill development. The participants of the study showed an improvement in their listening abilities with the auditory training/ learning provided. It must be understood that to maximize outcomes with an ABI, an auditory habilitation program for these recipients should (1) balance between communication approaches: 'auditory and oral/verbal language approach' and 'auditory and oral/verbal language approach with visual assistance' and (2) lay simultaneous emphasis on both 'bottom-up' -structured auditory training for development of listening abilities, from simple to more complex levels, and 'top-down' use of connected speech/ conversations to provide natural and holistic language and listening stimulation.

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. The habilitation programs for paediatric 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 population, are needed to confirm the utility of informal assessment like AuSpLan as an addition to formal testing.

Conclusions

Children with an ABI develop audition, speech, and language skills gradually over post implantation years. While formal assessment give good information about the development of the childand were encouraging, they 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 the development of children should not be underestimated.

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