31 Audiological Outcome with ABI

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31.1 Introduction

Cochlear implants (CI) have been widely used to provide auditory sensation in the profound sensorineural hearing loss. Despite their known effect on auditory perception, speech, and language development; in some cases (such as severe inner ear malformations (IEMs) and cochlear nerve deficiencies) CI has minimal or no effect on speech and language perception. At that point, auditory brainstem implants (ABI) can be considered for hearing restoration. In 2001, Colletti et al. reported the results of pediatric ABI patients for the first time, which showed environmental sound awareness and speech detection skills [1]. In the early 2000s, studies demonstrated that the ABI could be an option to improve speech and language development in these children [2–4]. Recently, ABI has started to be used with increasing numbers in many centers around the world with successful results on auditory perception skills in children. This chapter addresses the audiological outcomes of children with the ABI including auditory perception, language development, and speech intelligibility.

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31.2 Audiological Outcomes

Free field aided testing gives valuable information about the hearing level with ABI device. This test is routinely done in every follow-up after the initial device fitting in our implant group. The results from this test guide audiologists in changing the MAP parameters such as charge levels. Our pediatric ABI users with IEMs, who used their devices for a period of 1 month to 1 year after the initial fitting, have an average threshold of (average of 500, 1000, 2000, and 4000 Hz) 61 dB, whereas using the device for a period of 1–2 years brings average aided thresholds to 47 dB. Patients who used their ABI for a longer period (between 3 and 5 years) had better average aided thresholds of 44 dB. Finally, the group who used their ABI for more than 5 years had average aided threshold of 35 dB.

These are the mean average thresholds from all ABI users, but individual differences exist. Examples of individual differences in this sense were given in previous chapters with aided threshold examples of different ABI users. In fact, a user with high adherence to rehabilitation program and fitting sessions may reach the above mentioned aided threshold levels in a shorter time, and just the reverse may be observed for the ones who do not comply with our program. Existence of comorbid disorders has potential to worsen this even further.

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The number of active electrodes is another issue to be discussed in terms of outcomes. In a previous study of our group it was found that the number of active electrodes was not found to be related with closed and open-set speech recognition. In fact, active electrode percentage is 76% in our series, and this is not correlated with better aided thresholds or length of implant use. Better aided thresholds only have significant correlation with longer implant use period ($r = -0.571$, $p = 0.001$).

31.3 Auditory Perception Outcomes

From surgery to the first fitting, the families are anxious and confused during the waiting period. Therefore, it is necessary to inform families about the waiting period and the first fitting experience during preoperative period. After the first fitting, families often hope that their children will recognize the sounds immediately. To cope with frustration and hopelessness, it is necessary to explain the families that "hearing" is not "the transmission of voice through ears"; the brain plays a crucial role to recognize the meaning of sounds.

Although the child reacts to the sound during the ABI fitting, some children may not respond to speech sounds in everyday life. This is different from the cochlear implant (CI), because sound awareness abilities of CI users improve usually after the first fitting, and their listening behaviors are relatively clearer than ABI users. For this reason, after the initial tune-up, the family should be informed about how to monitor the children's responses to sounds and how to introduce the environmental sounds. Families often expect clear and typical listening behaviors. However, the first reactions of children with ABI include vague behaviors to the sound, and frequently there are unique behaviors in each child. For example, there may be relatively more clear responses with sounds, such as becoming quiet, slowing/accelerating movements, or less clear responses such as using gestures, shrugging their shoulders, and looking around. Moreover, parents should be informed that auditory perception process includes various development areas such as attention and memory skills. In this period, according to the information received from the family, auditory perception developments in daily life can be followed. Auditory perception skills of ABI users have been investigated with family reports, checklists, and assessment tools. The information about everyday listening behaviors can be obtained with questionnaires such as IT-MAIS [5], MAIS [6], FAPCI [7], and Little EARS [8]. These tools help parents to track their child's sound awareness behaviors. For further information, please refer to Chap. 8.

In the early 2000s, the promising results on auditory perception began to be published. Nevison et al. [9] shared their experience with 26 out of 27 adult patients who received auditory sensation during initial tune-up. According to follow-up outcomes, adult patients discriminated the basic temporal and spectral features of speech patterns. In one of the first studies, Colletti et al. [2] reported that environmental sound awareness and speech detection abilities improved after 1 year of experience. In 2009, Sennaroglu et al. published their preliminary results of 11 children with several cochlear malformations. All children were enrolled in the auditory-verbal therapy sessions and their parents chose aural communication. In their study, the auditory perception assessments were performed at 3 months' interval from first month to 1 year. Six children gained basic auditory perception skills such as identifying environmental sounds, recognizing and discriminating speech sounds after 6 months of experience with ABI. There was a regular increase in the MAIS scores of all children (Fig. 31.1). Same six children began to identify the Ling's Six Sounds at the end of a year. Only two children, who were diagnosed with attention deficit hyperactivity disorder (ADHD), scored inconsistently in Ling's Sounds Test. Sennaroglu et al. reported that children with additional handicaps such as ADHD, mental retardation, and developmental delay performed worse on

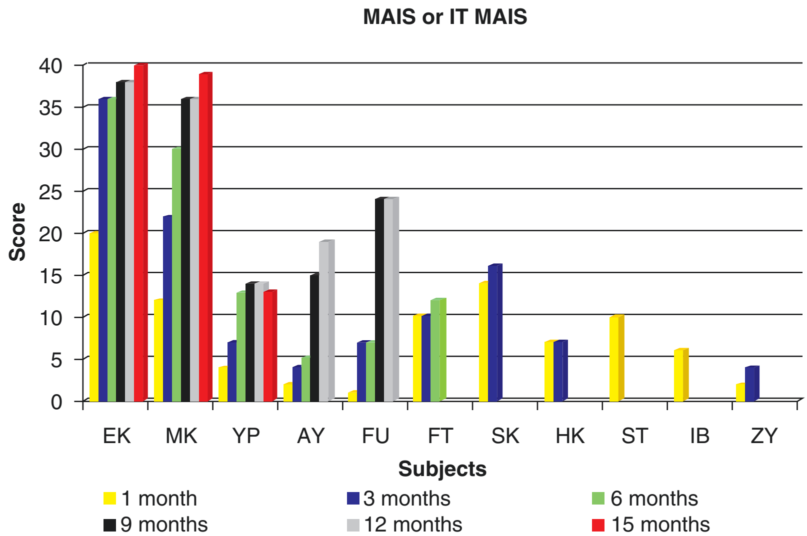


Fig. 31.1 IT/MAIS scores of ABI users in 15 months follow-up. (With permission from Otology & Neurotology)

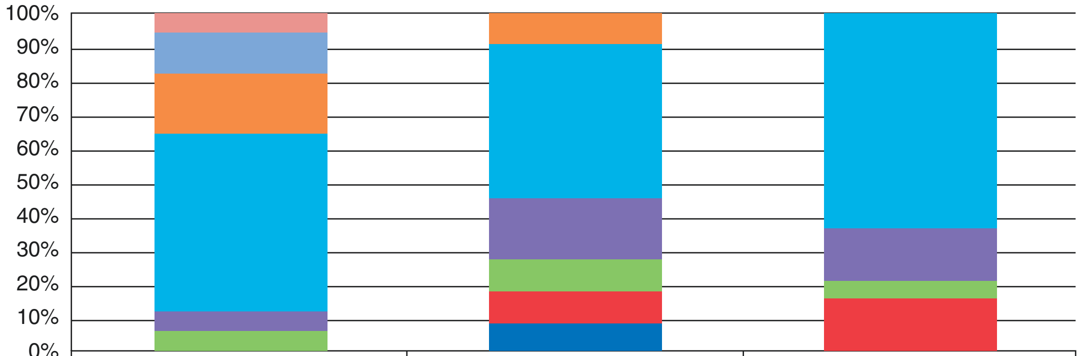
auditory perception tasks when compared to children with no additional handicap.

Findings by Choi et al. supported the developmental delay on auditory perception in children with additional handicaps such as mental retardation and blindness [10]. They believed that the reason for the delay could be the higher cognitive function, not associated with auditory sensation. This is because all eight children showed improvement in their auditory perception. In 2013, Hacettepe University pediatric ABI cohort's auditory perception outcomes were published and it included 39 patients with severe IEMs [11]. In their study, they reported the results of 29 children with ABI experience for 1.5 years. Most important finding was that children used their device regularly on daily basis, whether an additional handicap was present or not. In addition, majority of the children gained basic auditory perception skills. Eighty-six percent of children detected all six sounds in Ling's Sound Test; 75% of them recognized all sounds in the test, and 64% of them had MAIS scores between 30 and 40.

In 2016, Sennaroglu et al. reported their long-term results of 60 pediatric ABI users with complex IEMs [12]. Among 60 patients, 35 of them were followed more than 1 year. The rest of the patients that used ABI for less than 1 year were excluded from the study to eliminate the effect of inadequate experience with ABI. Auditory perception performance of the participants was assessed with CAP and they were divided into three groups according to their free field hearing thresholds. As demonstrated in Fig. 31.2, CAP scores were better with a better hearing threshold. Majority of the patients accumulated in category 5 which implies that they can understand common phrases without lip reading (Fig. 31.3).

Similarly, they compared the functional communication performance of the participants according to age at ABI surgery. Majority of the FAPCI scores were in the lowest tenth percentile, which reveals that children with ABI are worse performers compared to average CI users. There was no difference between the groups according to the age (Fig. 31.4). Moreover, they presented data that additional handicaps such as mental retardation, ADHD, and visual impairment had a negative impact on auditory perception performances of ABI users.

Wilkinson et al. reported that out of 9 children, who were enrolled in their ABI program, 4 of them completed the 1-year follow-up process. These children scored between 8 and 31 out of 40 points in IT-MAIS/MAIS, respectively [13]. They concluded that these scores demonstrated individual variability and slow progression as the children begin developing fundamental auditory skills. Moreover, they asserted that ABI outcomes



	25-40 dB	41-50 dB	>50 dB
8	5.9	0	0
7	11.8	0	0
6	17.6	9.1	0
5	52.9	45.5	57.1
4	5.9	18.2	14.3
3	5.9	9.1	4.3
2	0	9.1	14.3
1	0	9.1	0

Fig. 31.2 Distributions of CAP scores due to hearing thresholds of children with ABI. (With permission from Otology & Neurotology)

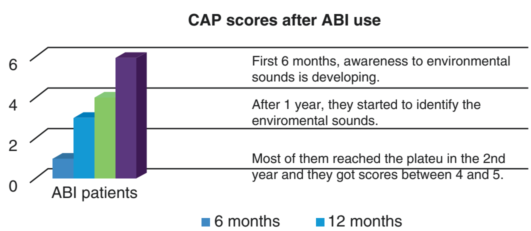


Fig. 31.3 CAP Scores of pediatric ABI users in 1 year period

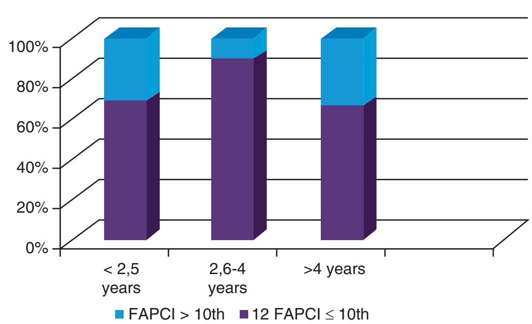


Fig. 31.4 Functional communication performances of children with ABI due to their age at surgery. (With permission from International Journal of Advanced Otology)

will occur along the mid-to-lower percentiles of CI patients due to longitudinal researches.

According to the literature [14, 15], it is evident that auditory perception of children with ABI showed individual variations. Despite the heterogeneity of the outcomes, all children used their device regularly and their auditory sensation was restored in different degrees. Conversely, children with additional handicaps showed slower development within the group. More intensive rehabilitation and integrative therapies should be considered in this latter group.

31.4 Speech Perception Outcomes

Speech perception development of pediatric ABI users was mostly assessed with closed-set pattern discrimination, word identification, and open-set sentence recognition tests. In cochlear implant (CI) technology, it is clear that speech perception

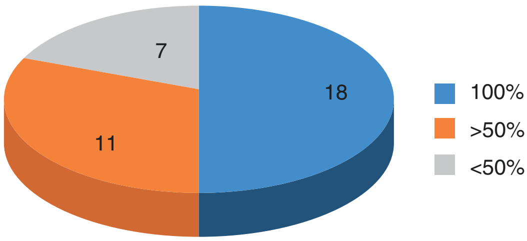


Fig. 31.5 Pattern perception performance. (With permission from International Journal of Advanced Otology)

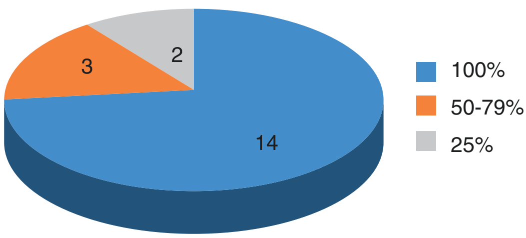


Fig. 31.6 Word identification performance. (With permission from International Journal of Advanced Otology)

ability of children with CI will improve steadily. In ABI technology, it is still unclear which factors are crucial for speech perception development. In this section, some of the factors that have impact on the speech perception outcome will be discussed.

One of the early articles on speech perception of ABI users investigated a child who was deafened in the perilingual period. Sanna et al. [16] shared their results of a 12-year-old female patient who underwent ABI after meningitis. The patient scored 100% sound identification, 90% recognition of bisyllable words, and 100% sentences recognition after 8 months. Later, Colletti et al. presented auditory perception performances of 14 ABI users [17]. Their initial findings indicated that three children began to recognize bisyllabic words and understand simple commands. While all children reached some degree of auditory sensation, none of them achieved open-set speech recognition yet. Case study by Eisenberg et al. provided information about speech perception development of 3-year-old boy who received ABI at the age of two [18]. After 1 year, he began to develop word identification in closed-set condition. Sennaroglu et al. reported 11 children's speech perception performances comprehensively [4]. Their findings showed that 4 of 11 children began to discriminate speech pattern, 2 of them had consistent word identification score of 24/24 at 9 months postoperatively (Figs. 31.5 and 31.6). Also, these children had improved their speech perception skills more rapidly than others. While four children continued to be struggling, only two children achieved open-set sentence recognition level after 15 months. They even began to use the telephone with familiar people. In the same study, children with no additional handicaps progressed steadily on speech perception tasks and showed that ABI provides a valuable opportunity for these children. In 2013, Sennaroglu et al. reported speech perception outcomes of 34 pediatric ABI patients after one and a half year [11]. The patients' sentence recognition performances improved and 10 of 29 patients scored 60–100 in auditory-verbal condition and 8 of them scored 20–100 in only auditory condition.

The first consensus meeting was organized on auditory brainstem implantation (ABI) in children and in non-neurofibromatosis type 2 (NF2) cases by Hacettepe University Implant Group in 2009. Health care professionals and scientists from ten centers who worked with pediatric ABI attended the meeting. At the end of the meeting, a consensus statement was published. One of the essential points on this statement was "from the results of different centers, it can be understood that it is possible to restore hearing perception in children with prelingual deafness with severe IEMs and cochlear nerve anomalies. In some selected cases, it also was possible to develop an open-set speech understanding. However, the family should be warned of different outcomes from this intervention so that their expectations should not be high. When compared with CI surgery, programming and rehabilitation of prelingually deafened children with ABI are much more labor intensive, and the results do not reach the level of CI users. On this basis, the candidacy assessment is much more detailed than in CI patients and requires more experienced staff. ABI is a viable option for children including prelingually deafened patients with IEMs and cochlear nerve hypoplasia/aplasia. ABI provides auditory perception in most patients. The potential for speech and language acquisition in the longer term depends on the age of implantation, the presence or absence of additional disabilities, and the other established factors seen in CI. It was concluded that open-set speech discrimination is possible in selected cases" [19].

In 2013, "Second Consensus Meeting on Management of Complex Inner Ear Malformations: Long-Term Results of ABI in Children and Decision Making Between CI and ABI" brought professionals from 20 centers in 11 countries. According to results from all centers, the pediatric ABI users could develop speech discrimination in closed-set to open-set condition [20]. At the time of consensus meeting, the largest case series had 35 children with at least 1 year of follow-up. In the cohort, 80% of children achieved scores above 50% with closed-set pattern discrimination task, while approximately 30% reached maximum scores. Additionally, 30% of patients scored above 50% with open-set sentence recognition test. The consensus report suggested that visual information should be used as a complementary element in auditory rehabilitation programs in pediatric ABI users. Follow-ups should be determined at shorter intervals than cochlear implantation and should be intensive.

31.5 Language Development Outcomes

While evaluating language development skills after auditory brainstem implantation, verbal language skills, as well as sign language and prevocalic development (gestures, mimics) skills, are equally important. For this reason, feedback from the family and the notes of the family and the experts working with the child are valuable for the evaluation of communication skills in daily life. In order to monitor the development, it is appropriate to request video and audio recording from families in different occasions. In this way, speech and communication analysis of the patient can be done more correctly.

At the end of the first 6 months, there is a significant increase in the vocalization of children. Their production has similar characteristics to babbling. In line with the development of pattern discrimination skills in auditory perception, they can produce sounds that are compatible with the patterns of words they try to imitate. The most common problem is the continuous repetition of the sounds produced by the child and the reduction of their intelligibility. For example, instead of "mama," they repeat "mamamama." In this period, children like to listen to their own voices, even the children who do not produce similar words like joy or anger are seen to make the sound plays.

In this period, experts should also consider to support the concept knowledge (such as colors, numbers) which is an important part of the language development. Additionally, another developmental area which is fine motor skill should not be ignored by focusing on the development of language and auditory perception. There is an individual difference in language performance in pediatric ABI patients due to chronological age, duration of ABI use, additional handicaps, and cognitive development. Sennaroglu et al. suggested that the main reasons could be the associated comorbidity due to additional handicaps such as attention deficit hyperactivity, slight mental retardation, and visual problems on language development delay [11]. They claimed that majority of the patients with limited improvement in performance have additional handicaps. Many studies have reported that supporting cognitive and fine motor skills can help support language development in children [21]. Eisenberg et al. [18] reported a 3-year-old boy whose language development reached the level of 2-year-old children. Although he had a normal intelligence, he had difficulties in completing structured tasks due to attention deficits.

According to the studies, language development skills of children with ABI are not compatible with their typically developing peers, but they are more prone to show language development compatible with the time they begin to hear [22]. Depending on the individual differences of children, the time, when the child is ready for standardized language development tests, shows variation. When language skills are examined in two subtests (receptive and expressive language skills), receptive language is slightly better than the expressive language [4]. The long-term follow-up outcomes of language development showed that the language skills of the patients have entered into a stagnation period [12]. The language development in the first 2 years of ABI surgery is beyond the duration of ABI use. In 2009, the language scores of 15 patients were above the duration of ABI use in means of language parameters as age equivalent scores (Figs. 31.7 and 31.8). However, when this same group of patients is examined almost 5 years later, only 4 of 20 are above the line (Figs. 31.9 and 31.10). It is assumed that the rate of language development was much faster in the first 2 years, and although it still continues, the pace declines afterward.

Manual communication options (such as sign language, coded language, cued speech) should be recommended to parents' of children with ABI to support their communication skills. It is observed that communication skills increase with the support of sign language, behavioral problems decrease, provide children to follow the conversation with more than one person and in noisy environments [14, 22]. These findings were supported by the first consensus statement as "Auditory-verbal therapy in these children, where only auditory stimulation is conveyed, may not be as efficient as in children who are using CI. Total communication and speech reading also should be encouraged to convey more linguistic and language information to these children. In this method, speech reading assumes considerable importance as a source of information, whereas tactile and motor kinesthetic stimulation provides supportive avenues for spoken language acquisition. In addition, the involvement in speech reading training programs has a positive effect on postoperative perceptive and expressive linguistic skills" [19].

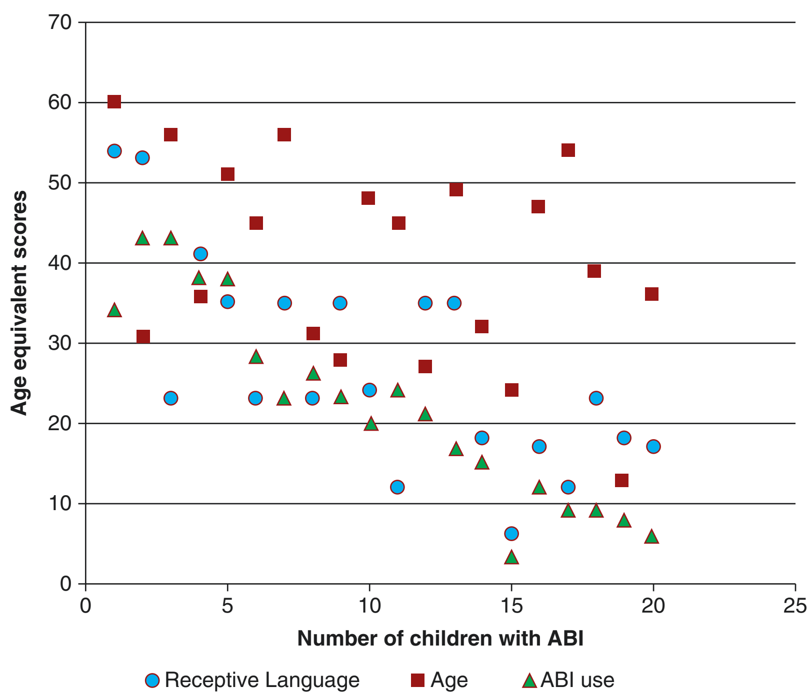


Fig. 31.7 Receptive language performance in 2009 study. (With permission from Otology & Neurotology)

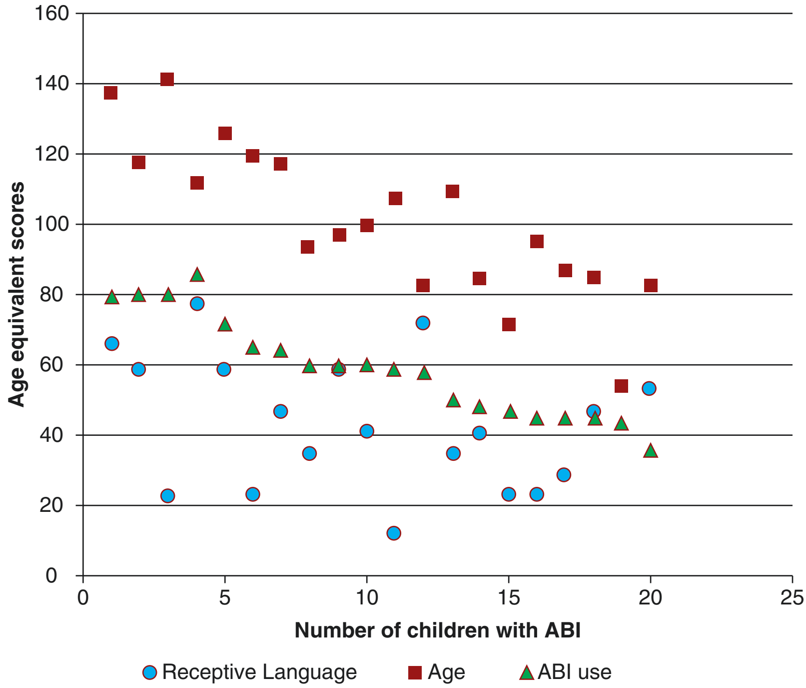


Fig. 31.8 Receptive language performance of the same patients in 2013 study. (With permission from Otology & Neurotology)

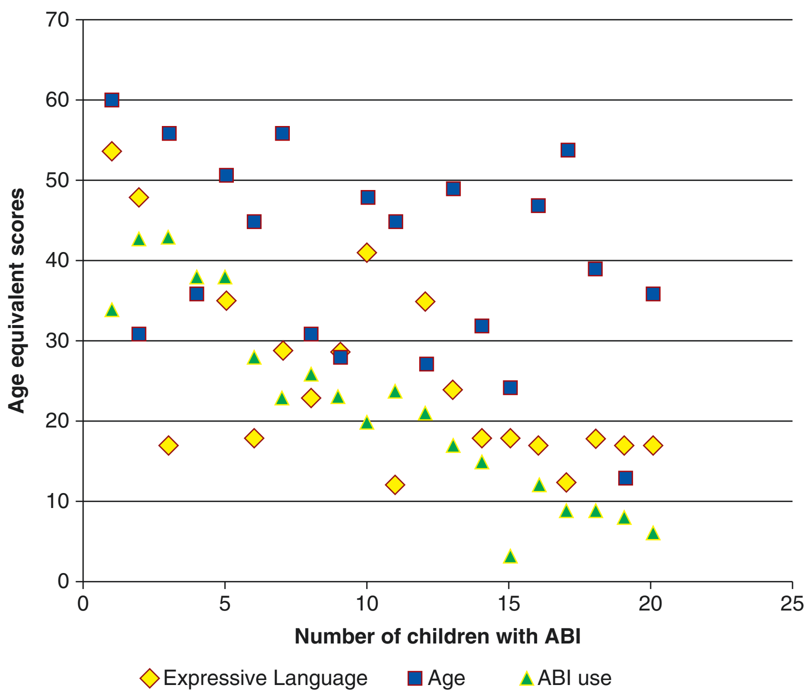
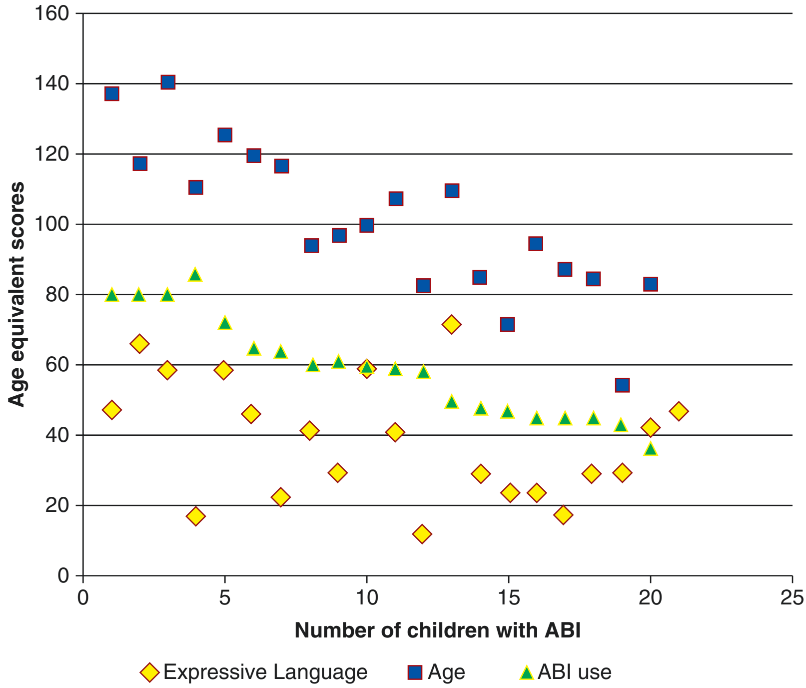


Fig. 31.9 Expressive language performance in 2009 study

Fig. 31.10 Expressive language performance in 2013 study



31.6 Speech Intelligibility Outcome

In addition to expressive language skills, there are difficulties in developing speech intelligibility which is low in pediatric ABI patients [12]. Speech intelligibility is reported to be one of the most challenging skills in daily life in the assessments and information obtained from the family [22]. According to Eisenberg et al. parents reported that their children's speech remains unclear, especially for unfamiliar listeners [14]. Therefore, they may need additional cues such as sign language for communication. Further studies are needed in the literature on the difficulties of speech intelligibility in this population.

31.7 Recent Results of Pediatric ABI Patients

Between June 2006 and September 2018, 124 children with complex IEMs received an ABI by Hacettepe University Hospital Implant Team. Five of these are revision due to device failure. The results of 84 primary pediatric ABI patients who have been using their ABI for more than a year were analyzed and presented in 15th The International Conferences on Cochlear Implants and Other Implantable Auditory Technologies in Belgium. Inclusion criteria for this study include: (1) age younger than 18 years, (2) monolingual children, (3) only unilateral ABI users, (4) at least 1-year ABI experience. Children with additional difficulties were not excluded from this study, only one child with autism who was not enrolled in the sessions regularly was excluded.

Fifty-nine percent of the patients were female. Mean age at ABI surgery was 34.54 months old (range: 12–96 months old, SD = 18.52). Of all patients, 26% had additional difficulties such as CHARGE syndrome, Goldenhar Syndrome, ADHD. Seventy-six percent of these children are using auditory-verbal communication mode and the rest have chosen total communication.

In this study, average MAIS score was 26.50 (range: 5–40, SD = 10.83). There is a significant relationship between MAIS scores and age of

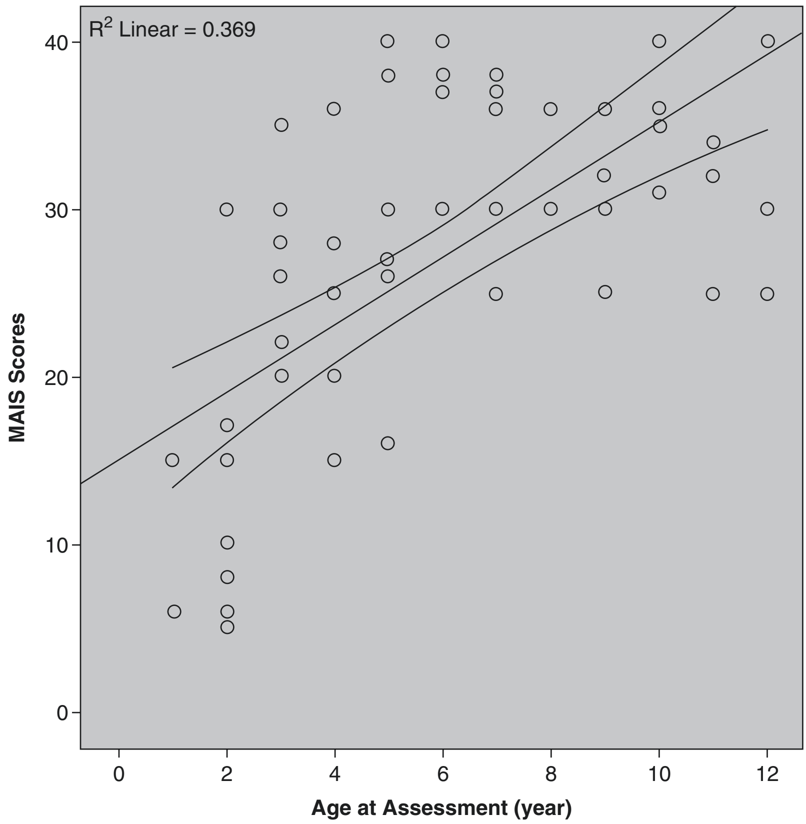


Fig. 31.11 MAIS performances due to age at assessment

evaluation (Fig. 31.11). Although there was a linear development due to chronological age in MAIS performances, no significant relationship due to age at ABI surgery was found.

Out of 84 children who met the inclusion criteria, we determined that approximately half of the participants completed the ability to distinguish the closed-set pattern. We found that 17% of the children could do more than half of the test and 36% of them continued to improve this skill. While the difficulty in "word discrimination" testing increased, we wanted them to find the word that has the same number of syllables; 22% completed this skill, 31% completed more than half of the test, and 46% still continue to improve this skill. The next stage is the open-ended sentence recognition phase where clues are eliminated. At this stage, 16% of children recognized the sentences in daily life, 24% of them are just starting to gain this skill, and 63% of them have not yet reached this level in auditory perception.

When we evaluated long-term language development results of children with ABI, the gap between chronological age and language age persists. In contrast to the difference between chronological age and language age, the gap between the age equivalence of language performance and the duration of he/she begins to hear (hearing age) decreases. We found that language development of children who started to use ABI in the early period was similar when they started to hear. There was no statistically significant difference between the language development of the participants who had started using ABI before and after the age of 3 years. Therefore, the following long-term results are important for determining whether the gap would be opened or closed (Figs. 31.12 and 31.13).

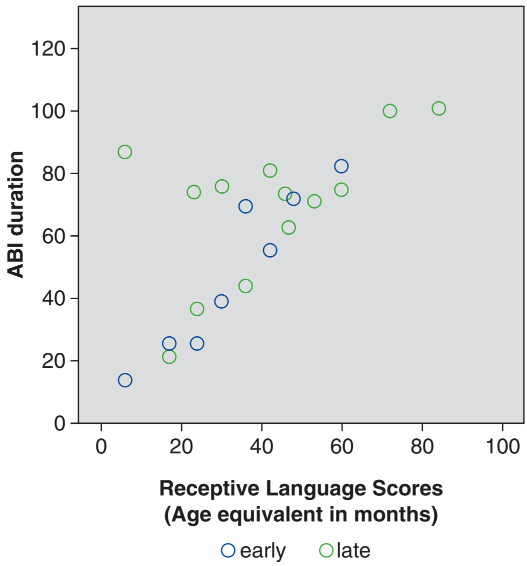


Fig. 31.12 Receptive language scores of ABI users due to duration of ABI use

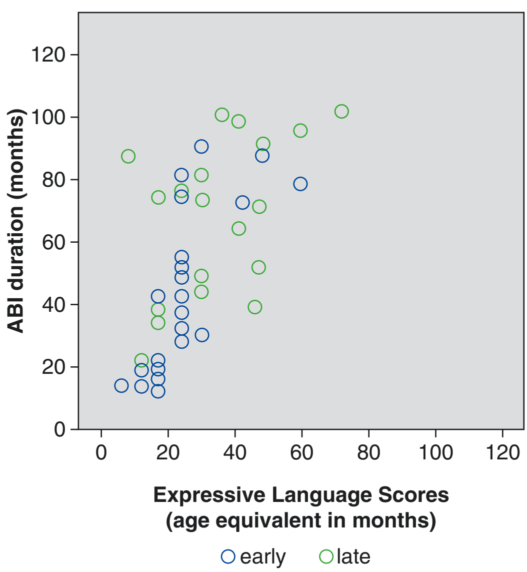


Fig. 31.13 Expressive language scores of ABI users due to duration of ABI use

31.8 Educational Settings

Children who are being considered for ABI candidacy should be evaluated in terms of cognitive skills and learning behavior. Their participation in educational tasks is decisive for postoperative progress and building of a therapy program. The assessment process involves their communication behaviors such as eye contact, speech reading, turn-taking, initiating, and sustaining joint attention, following directions, etc. [20]. These nonverbal forms of communication reflect the child's ability to structure a particular communication method [20]. In 2018, 47 of 75 children with ABI at school age, only 9% of them attended the school of Deaf. All children were enrolled in Individualized Education Programs in local centers. Majority of the children used total or verbal communication. Further studies need to be done to determine which factors have a significant impact on the choice of school settings (Figs. 31.14 and 31.15).

Factors that can influence auditory perception and language outcomes

- ✓ Length of auditory deprivation
- ✓ Age at surgery
- ✓ Etiology of deafness
- ✓ Additional handicaps
- ✓ Surgical issues
- ✓ Mode of communication
- ✓ Educational settings
- ✓ Motivstion and participation of parents
- ✓ Post-operative hearing thresholds
- ✓ Training options and frequency

Fig. 31.14 Factors that can influence auditory perception and language outcome in pediatric ABI users

Standard protocol includes in pediatric ABI users

- ✓ Reports about child's listening behaviors and use of ABI
- ✓ Comment of the therapist
- ✓ Behavioral audiometry
- ✓ Check of device function
- ✓ Measurement of electrodes
- ✓ NRT recording

Fig. 31.15 The parameters should be included in standard assessment and follow-up protocol in ABI

31.9 Conclusion

An auditory brainstem implant is an auditory prosthesis that provides acceptable and effective treatment option for pediatric population with complex IEMs. Auditory brainstem implants have enabled children, who show no or inadequate benefit from cochlear implants, to develop speech perception and language skills. It is evident that the majority of the children gain auditory sensation with ABI. Therefore, the speech perception and language outcomes varied widely.

Although children with ABI demonstrated improved speech perception ability with auditory-verbal approach, visual cues and sign language should be offered to enhance their communication skills. Even children, who achieved open-set sentence recognition, need visual cues in difficult listening environment.

It is recommended that the family and the professionals working with the child are informed carefully about the (re)habilitation objectives. More realistic targets keep therapists and parents on the track to support children with ABI. The motivation and participation of the family and the child in the rehabilitation process are very important.

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