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Young cochlear implant users' auditory development as measured and monitored by the LittlEARS® Auditory Questionnaire: A Turkish experience

Julie Kosaner [a,*], Samime Sonuguler [b], Levent Olgun [b], Edda Amann [a]

[a] MED-EL Worldwide Headquarters, Fuerstenweg 77a, 6020 Innsbruck, Austria
[b] Ministry of Health Research and Education Hospital, Bozyaka, Bahar Mh., 35170 Izmir, Turkey

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ABSTRACT

Objective: This study aims to assess the usefulness of the LittlEARS® Auditory Questionnaire (LEAQ) in determining the audiological development of Turkish children who have received a cochlear implant.
Methods: 20 children received a cochlear implant before their 3rd birthday. Each child's progress was evaluated with the LittlEARS® Auditory Questionnaire at first device fitting and then at 3-month intervals for 2 years. Scores were compared with the age-related norms established by hearing children.
Results: All children showed a significant increase in LittlEARS® Auditory Questionnaire scores over time. Nearly all children showed a growth in auditory skills similar to that of hearing children. Children without additional needs showed more development than did children with additional needs.
Conclusions: The LittlEARS® Auditory Questionnaire is useful for monitoring the audiological development of young children with a cochlear implant. Confirmation that a cochlear implant user is achieving typical auditory milestones serves to boost parental morale during a child's pre-verbal stage when parents may be anxious about their child's ability to talk. The questionnaire could also be useful as an early warning system. Poor scores likely indicate that something is impeding the child's development. This should prompt professionals to try to identify the impediment, whether technical, medical, social or educational and, possibly, eliminate/mitigate its effects while the child is still in his/her critical development stages.

1. Introduction

Cochlear implants (CI) give young hearing-impaired children access to sound, enabling auditory development, a pre-requisite for linguistic development and some forms of cognitive development. Auditory behaviour is age-dependent – it changes over time – and thus so is linguistic and other forms of development. Given the challenges of hearing with a CI, it is essential to assess and document the children's audiological development to ensure they are getting the sound they need to develop along age-expected learning curves. In prelingually deaf children, this is especially important to do at least before their 3rd birthday, as a vast body of research has shown that children's ability to acquire language irretrievably declines after this time [1–4].

An effective assessment tool for auditory development is available for young children. The LittlEARS® Auditory Questionnaire (LEAQ) [5] is a validated, language-independent tool for assessing preverbal and early verbal auditory behaviour in normal-hearing children up to 24 months of age [6]. It examines children's responses to sound and their vocal-verbal productions, and relates them to age-dependent milestones of auditory behaviour. The LEAQ has 35 yes/no questions, to be answered by parents at home after observing their child in his/her normal activities, i.e. a testing situation does not need to be artificially created and the child need not know he/she is being evaluated. Examples are given to clarify the information the questionnaire is seeking and thereby increase the objectiveness of the questionnaire. The tested child's score can be compared with the norm curve showing expected and critical values to check if the child's progress is typical. One "point" is awarded for each "yes" answer, 0 points for "no." The norm curve of the LEAQ was created from the scores of 218 normal hearing German native-speaking children but has been adapted into 15 additional languages and is language-independent [6]. We translated the LEAQ into Turkish (and back translated to verify

* Corresponding author at: MEDers İşitme ve Konuşma Merkezi, Söğütlüçeşme Caddesi No: 102, 34714 Kadıköy, İstanbul, Turkey. Tel.: +90 216 348 09 39; fax: +90 216 348 02 43.
E-mail address: julie.kosaner@meders.com.tr (J. Kosaner).

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accuracy) and used it without first validating it on normal-hearing Turkish-speaking children.

The LEAQ can also be used on children who are hearing-impaired (HI) to evaluate (or confirm) the need for intervention and to document progress after the provision of a hearing device [7]. With children who are HI, the "hearing age" (i.e. length of device experience) and not the chronological age is used for comparison with norms. The LEAQ can thus be used on HI children with up to 24 months of hearing age.

This prospective study aims to document the auditory development of early implanted children and compare it to that of hearing children.

2. Methods

2.1. Subjects

Our subjects were 20 children implanted before the age of 36 months in the CI centre of the Izmir Training and Research Hospital in Izmir, Turkey from 2007 to 2009. All children were bilaterally profoundly deaf according to their auditory steady state response data (mean hearing loss across 500, 1k, 2k, and 4k Hz). All children derived insufficient benefit from the at least 1 hearing aid they wore, in accordance with Turkish law, for at least 3 months prior to implantation. Prior to implantation, none of the children used sign language only 3 (#s 8, 11, 12) used some spoken words (between 1 and 10). 2 children (#s 13 and 19) had less developed eye contact, turn taking, and information sharing skills than the remaining 18 children who had well-developed communication skills. We would like to note that the possible presence of additional needs was *not* an exclusion criterion because this study aimed to evaluate the "real world" (in which 30–40% of CI users have an additional need [30,31]) application of the LEAQ, it was not a study to validate the LEAQ on CI users. The above data show that the 20 study children were similar to each other in terms of hearing loss, language development, and the benefit they received from the hearing devices they has used prior to CI. The differences observed in their LEAQ scores at 24 months post-CI would therefore seem unrelated to their pre-implant hearing loss or their use of amplification in the non-operated ear.

All children were unilaterally implanted with the PULSARCI[100] and used the OPUS 2 audio processor with FSP (Fine Structure Processing) sound coding strategy (MED-EL, Innsbruck, Austria). Surgeons used a standard posterior tympanotomy approach, inserting the electrode in 16 children via the round window and 4 via cochleostomy.

All audio processors were optimally fitted by an experienced clinician. The children's fitting parameters were rated ideal, satisfactory, or less than ideal. Ideal was defined as having 11 or 12 active electrodes, maximum comfortable levels (MCL) based on electrically elicited stapedius reflex threshold measures (eSRT) or through observation of auro-palpebral reflexes (APR), pulse durations < 40 ms, 1 or more fine structure (FS) channels, and a rate > 1200 pps. Satisfactory was defined having 9–10 active electrodes, no eSRT, some APRs, some pulse durations > 40 ms, 1 or more FS channels, and a slower rate around 1000 pps. Less than ideal was defined as having 8 or fewer active electrodes, no eSRT or APR, all pulse durations > 40 ms, no FS channel, and a slow rate < 1000 pps. 13 children had ideal fitting parameters, 5 had satisfactory, and 2 had less than ideal. Not all CI users can be ideally fit because of the uniqueness of their auditory system. The variation in this study group's fitting ratings is similar to those found in any group of CI users. Less than ideal fitting parameters would likely affect outcomes in terms of auditory performance, this however was not statistically pursued due to insufficient subject number for each fitting group and the presence of confounding factors like additional needs and age. The mean age at first fitting was 25.9 months (range 13–35 months) (Table 1.).

After implantation, all children were entitled to go to a Ministry of Education monitored habilitation centre for 2×45 min individual sessions per week with a teacher of the deaf, educational audiologist, child development specialist, nursery class teacher, or psychologist. All of the habilitation centres used a natural auditory oral approach to language development.

All parents signed a protocol explaining the study, how the results would be used, and what study participation would entail. They were fully informed that they could withdraw their child at any time without repercussion.

LEAQ data for normal hearing children were taken from the established age-dependant norm curves. No additional normal hearing children were involved in the present study.

2.2. Comparing auditory development (LEAQ) of early implanted children with that of typically developing children

The children were evaluated with LEAQ tests at first fitting and then in regular 3 month intervals until 24 months after first fitting. If a child scored the maximum of 35 before 24 months, we deemed

Table 1
Subject demographics: for fitting rating, 2 = ideal, 1 = satisfactory, and 0 = less than ideal.

Subject #	Sex	Age at first-fitting (months)	Aetiology	Fitting rating	Additional comments
1	M	24	Hereditary	0	
2	M	20	Unknown	2	
3	F	25	Hereditary	2	
4	F	28	Ototoxic medication	2	Motor delay
5	F	32	Hereditary	2	
6	M	32	Unknown	1	
7	F	18	Unknown	0	Thin CN 8
8	M	31	Hereditary	2	
9	M	20	Hereditary	1	
10	F	34	Waardenburg synd.	2	
11	F	26	Unknown	2	
12	F	25	Hereditary	1	
13	M	22	Premature	1	Motor and cognitive delay
14	M	15	Unknown	2	Chronic otitis media
15	F	34	Seizure at 6 months	1	
16	F	13	Unknown	2	
17	F	22	Hereditary	2	
18	F	32	Congenital rubella	2	
19	M	35	Hereditary	2	General motor delay, autistic tendencies
20	M	29	Hereditary	2	Both parents congenitally deaf

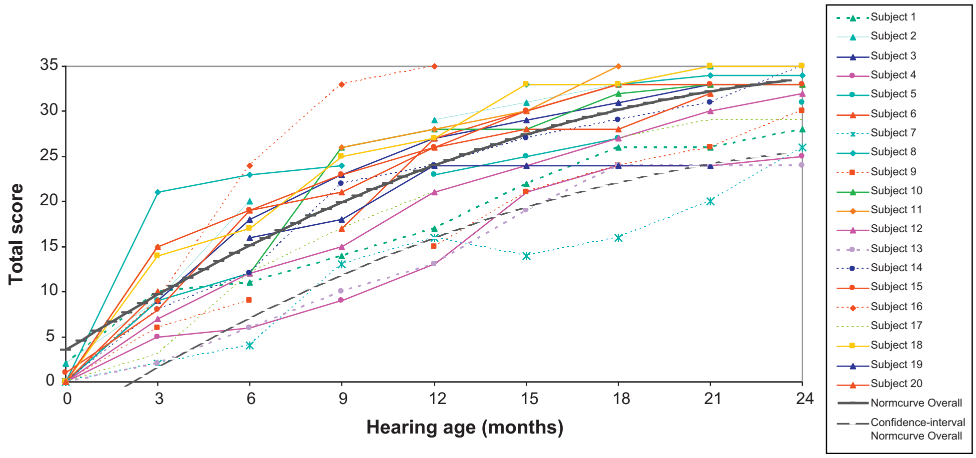

Fig. 1. Individual results of 20 CI-children – gradient from first fitting to 24-month testing.

it unnecessary to continue LEAQ testing and included his/her final score as endpoint data. LEAQ tests were performed via interview.

We compared the LEAQ scores in terms of hearing age for the implanted children with the norm curve of normal hearing children [6]. We used one-way repeated measure ANOVA with time as a factor to look for a significant increase in CI users' LEAQ scores over time. For the ANOVA, Mauchly's test of sphericity was applied. If sphericity could not be assumed, we used a Greenhouse–Geiser correction as part of the ANOVA. We performed post hoc pair-wise comparisons adjusted with the Holm–Sidak method to look for a significant increase in LEAQ scores between consecutive test intervals. We regarded $p \leq 0.05$ as significant for all tests.

2.3. Age at implantation

Although age at implantation is a very important factor effecting outcomes, we have not here analysed it as a variable. This is because the current debate on when best to implant a child mainly focuses on whether it is more beneficial to implant children before their first birthday [8–12] or wait until they are 1–2 years old [13,14]. In Turkey, at the time of the study, implantation under 2 years (except in the case of meningitis) was not very common and so our study population does not allow us to address this question. Further, our study aim was to demonstrate that the LEAQ benefits a variety of children, not those of carefully selected demographic groups.

3. Results

3.1. Subjects

1 child (# 7) had inadequate hearing due to a thin CN 8, although his/her implant was correctly placed and functioning. She received a 2nd (bilateral) implant 17 months after having received the 1st CI; however, even with the 2nd implant her audition was less than optimal because of non-auditory stimulation. This child and child # 1 had a less than ideal fitting rating. Despite having supportive parents and attending a rehabilitation centre consistently, both scored around critical level on the LEAQ at 24 months post-activation (# 7 had 26 points, # 1 had 28), as was expected. 19/20 children continued to use a hearing aid (HA) in the non-implanted ear after CI to stimulate the ear and possibly provide some useful low frequency information. Only 1 child (# 14) did not wear a hearing aid on non-operated ear. As the children in this study had profound bilateral hearing loss, using a HA in the other ear is unlikely to have affected outcomes on the LEAQ.

In addition to twice weekly sessions at a rehabilitation centre 10 children (#s 1, 2, 6, 7, 8, 11, 13, 15, 18, 20) attended the CI centre for parent guidance on a month basis, the other 10 children attended the CI centre for parent guidance less regularly. One child did not attend a rehabilitation centre because he/she lived in a remote area.

3.2. Correlating CI children's LEAQ scores with age-established norm curves

CI users had zero or near zero LEAQ scores at the start of the study but showed a statistically significant increase over the 24 month evaluation period, according to the result of a repeated measure ANOVA (F (2.2, 21.7) = 142.92, $p < 0.001$). Post hoc pair-wise comparisons adjusted with the Holm–Sidak method generally confirmed this statistical significant improvement ($p < 0.001$–0.050) (Fig. 1).

CI users' mean LEAQ score was 31.1 (range 24–35) at 24 months (hearing age), which lies between the critical score of 27 and the expected score of 33 for typically developing hearing children at 23–24 months (chronological age). While 4 children (#s 4,7, 13,and 19) had a LEAQ score of less than 27 after 24 months of device use, 4 children (#s 2, 11, 16, and 18) achieved a LEAQ score of 35 before 24 months, one of whom, # 16, had a LEAQ score of 35 at just 12 months hearing age (see Table 2).

Looking at the individual data, the rate of progress across children varied: some children made large improvements across certain intervals while others made little progress; e.g. child # 8's score increased from 0 to 21 from the 1st to the 2nd test interval, while children #s 7 and 13 scores increased from just 0 to 2 in the same interval.

The increase in LEAQ scoring was not significant between 3-month and 6-month testing ($p = 0.088$), 12-month and 15-month testing ($p = 0.104$), 18-month and 21-month testing ($p = 0.318$), 18-month and 24-month testing ($p = 0.335$), and between 21-month and 24-month testing ($p = 0.833$).

4. Discussion

4.1. Comparing the LEAQ results of children with CI with those of their hearing peers

24 months after implantation the children's auditory development was similar to that of normal-hearing children. The children with a CI had a mean LEAQ score of 31.1, slightly below the norm curve, 33, but safely above the critical score, 27, on the border between normal and poor auditory development. If we exclude the

Table 2
LEAQ results at all tested intervals. FF = first fitting. M = month interval.

Subject #	FF	3 m	6 m	9 m	12 m	15 m	18 m	21 m	24 m	Additional comments
1	2	10	11	14	17	22	26	26	28	Less than ideal fitting parameters
2	2	9	20		29	31	33	35		Supportive parents
3	0	9	18	23	27	29	31	33	33	
4	0	5	6	9	13	21	24	24	25	Unsupportive parents, missed many appointments
5	0	9	12		23	25	27		31	
6	0	10		17	27	30	33	33	33	
7	0	2	4	13	16	14	16	20	26	Poor hearing with her CI. Received bilateral implant 17 m after 1st implant
8	0	21	23	24	31	33	33	34	34	
9	0	6	9		15	21	24	26	30	
10	0		12	26	28	28	32	33	33	
11	0	15		26	28	30	35			Supportive parents
12	0	7	12	15	21	24	27	30	32	
13	0	2	6	10	13	19	24	24	24	Developmental delay, unsupportive parents
14	0	8	12	22	24	27	30	31	35	
15	1	8	19	23	26	30	33	33	33	
16	0	9	24	33	35					Supportive parents
17	0	3	12	17	21		27	29	29	
18	0	14	17	25	27	33	33	35	35	Supportive parents
19	0		16	18	24	24	24	24		Autistic tendencies, badly behaved, missed many appointments
20	0	15	19	21	26	28	28	28	32	Missed many appointments, both parents deaf, limited input of spoken language

scores of the 3 children with additional needs (#s 4, 13, 19), the mean score rises to 32.29.

These findings are in line with previous studies [7,15–17]; all of which found that many children implanted with a CI before their 3rd birthday are able to achieve similar results as their hearing peers. It should, however, always be taken into account that this does not mean children with a CI have the same audiological abilities as children with NH, just that with cochlear implantation and habilitation, many congenitally deaf children are able to meet the same developmental benchmarks.

Although the amount and quality of parental support is known to affect audiological development [14,18,19], it is difficult to objectively quantify, however, we considered parents "supportive" when they (1) were regular and punctual in rehabilitation session attendance, (2) keen to interact with their child and participate during the rehabilitation sessions, and (3) the quantity and quality of shared activities with their child at home that they were able to report. The less the parents did the above activities, the less supportive we regarded them. During the study we noted that children #s 1, 2, 7, 8, 11, 14, 15, 16, and 18 had especially supportive parents. These 9 children had a mean LEAQ score of 32.89. If you exclude child # 1 (fewer active electrodes, subjectively determined MCLs, no FS channel and a relatively slow processing rate) and child # 7 (thin 8th CN nerve and reimplantation) the mean rises to an impressive 34.57. We cannot here assess the effect of unsupportive parents, as only children with an additional need had parents we regarded as unsupportive.

Unsurprising, the children with an additional need (#s 4, 7, 13, and 19) performed poorer (mean 24.75 at study end) than did the other children. Previous studies have also noted this slower development [14,20,21]. 3 of these 4 children also had parents we noted as unsupportive; a difficult combination for a child to overcome. Indeed, we felt that child # 4, despite his/her additional needs, would have developed well if his/her family had been more supportive.

Other factors known or thought to influence the audiological development of children with a CI were not studied. Factors such as parental socioeconomic level [4,22], parental IQ and education level [19,23], family size [24], and child intelligence [23,25,26] were out of the present study's scope. Hopefully future studies can examine these variables in detail.

We believe that the real value of the LEAQ, as our results show, is not only that it confirms that individual children are progressing according to age-expected norms, but that it clearly shows that some children are not. Confirmation that a child is progressing as expected during the time before a child actually produces words helps to sustain parental morale, encouraging parents to interact with their child in a "normal" and positive way. Poor LEAQ scores (children #s 1, 4, 7, 13, and 19) indicate the possible need for an additional intervention. This may be as simple as trouble-shooting an external device problem or providing a more appropriate audio processor programme. In rare cases, the problem may be much more serious and may involve a revision or re-implantation of the electrode array. If the child can hear well but is not showing typical auditory progress, a review of habilitation services and the family environment is indicated. The situation may be ameliorated by provision of more frequent, more intensive family-centred habilitation. Poor LEAQ scores may prompt early identification of an additional need (or needs) giving rise to early involvement of professionals with expertise in the child's additional need area. A child with additional needs may benefit from habilitation materials and methods specifically designed for children with that need, as has been suggested [27]. Early discovery of additional needs may help parents to develop more realistic expectations for their child, saving them from some of the stress, disillusionment, and even anger that can follow their child's inexplicably poor performances [28,29]. Detailed documentation of a child's auditory development as provided for by the LEAQ allows professionals to effectively share information with other professionals such as service providers.

The children in our study had a mean age of 25.9 months at first fitting, 10.1 months older than the hearing-impaired children the LEAQ was validated on [7]. To make a true comparison between the auditory development of CI users and that of normally hearing children, the CI users would need to be of the same age; however, as most clinics implant children older than 12 months, especially in Turkey, a direct comparison is difficult. However, a study comparing the auditory development of very young implanted children with hearing children would lessen the effect of higher degrees of maturation and reveal a clearer picture of auditory development with a CI.

5. Conclusions

Most profoundly deaf children implanted before their 3rd birthday will be able to achieve the same auditory milestones at the same rate as normal hearing children do, if these implanted children do not have additional needs, when they are provided with the same opportunities as hearing children, their auditory abilities should allow them to develop spoken language. The LEAQ is a useful tool to monitor children's pre- and early verbal auditory behaviour to ensure its development is consistent with age-expected norms. This is especially true in children with a CI as their audiological development is more subject to impediments and may thus be more fragile. Low LEAQ scores can quickly alert parents and professionals to possible hearing problems. Such early alert could lead to the provision of better adapted audiological therapy or even early detection of a yet undetected concomitant disorder; a detection which would be likely to benefit both parent and child.

Conflicts of interest

Authors Julie Kosaner and Edda Amann, and medical writer Michael Todd are all MED-EL employees. The study was, however, not funded by MED-EL. Otherwise, the authors report no conflicts of interest.

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