TOWARDS A UNIFIED METHOD OF ASSESSING EARLY AUDITORY DEVELOPMENT AFTER COCHLEAR IMPLANTATION: MEASURING RELATIVE DELAY

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Abstract

Current research on auditory development in children with profound sensorineural hearing loss emphasizes the importance of providing effective auditory stimulation as early as possible, preferably before 12 months of age. Cochlear implants have been identified as the most effective method for delivering sound to the auditory system in these children. To minimize neurological consequences of congenital profound sensorineural hearing loss it is vital to monitor all stages of auditory development right from the moment of implantation. The LittlEARS Auditory Questionnaire, by measuring hearing ability in children up to 24 months of age, facilitates such an assessment. In this paper multiple approaches to reporting results of the LittlEARS are reviewed. Two difficulties with interpreting the results from cohorts of cochlear implanted children are identified, one related to variability of the results and the second to the age of the child at intervention. To overcome these difficulties a method of calculating the "relative delay" in auditory development is proposed. The method should facilitate between-group comparisons in single or multicenter studies, as well as improve meta-analysis of data in evidence-based practice.

Keywords: children • cochlear implant • auditory development • parental questionnaires • congenital profound sensorineural hearing loss

W STRONĘ UJEDNOLICONEJ METODY OCENY WCZESNEGO ROZWOJU SŁUCHOWEGO PO IMPLANTACJI ŚLIMAKOWEJ: POMIAR WZGLĘDNEGO OPÓŹNIENIA

Streszczenie

Aktualne badania rozwoju słuchowego dzieci z obustronnym głębokim niedosłuchem odbiorczym wskazują na konieczność zapewnienia skutecznej stymulacji słuchowej tak wcześnie, jak to możliwe, najlepiej przed ukończeniem przez te dzieci 12 miesiąca życia. Zastosowanie implantów ślimakowych (CIs) uznawane jest obecnie za najskuteczniejszą metodę dostarczania dźwięku do układu słuchowego u tych dzieci. Aby zminimalizować neurologiczne konsekwencje wrodzonego głębokiego niedosłuchu zmysłowo-nerwowego u dzieci, konieczne jest monitorowanie wszystkich etapów ich rozwoju słuchowego od momentu wszczepienia implantu. LittlEARS – kwestionariusz przeznaczony do oceny rozwoju słuchowego u dzieci do 24 miesiąca życia – ułatwia taką ocenę. W niniejszym artykule dokonano przeglądu różnych metod raportowania wyników LittlEARS. Zidentyfikowano dwie trudności z interpretacją wyników dzieci korzystających z implantów ślimakowych: jedną związaną ze zmiennością wyników, a drugą – z wiekiem dziecka w momencie interwencji. Aby przezwyciężyć te trudności, zaproponowano metodę obliczania tzw. względnego opóźnienia rozwoju słuchowego. Metoda ta powinna ułatwić porównania między grupami w badaniach jedno- lub wieloośrodkowych, a także umożliwić metaanalizę opublikowanych danych.

Słowa kluczowe: dzieci • implant ślimakowy • rozwój słuchowy • kwestionariusze wypełniane przez rodziców

Key to abbreviations	
AAST	Adaptive Auditory Speech Test
ABEL	Auditory Behavior in Everyday Life
CAEPs	cortical auditory evoked potentials
CHILD	Children's Home Inventory for Listening Difficulties
CI	cochlear implantation

Key to abbreviations	
CIs	cochlear implants
CPA	Conditioned Play Audiometry
ELF	Early Listening Function
ESP	Early Speech Perception (test)
FAPI	Functional Auditory Performance Indicators
IT-MAIS	Infant–Toddler Meaningful Auditory Integration Scale
LEAQ	LittlEARS Auditory Questionnaire
LIP	Listening Progress Profile
MAIS	Meaningful Auditory Integration Scale
NH	normal hearing
NU-CHIPS	Northwestern University–Children's Perception of Speech (test)
P1	first positive peak
PEACH	Parent's Evaluation of Aural/Oral Performance of Children
PICO	patient, intervention, comparator, outcome
PSI	Pediatric Speech Intelligibility
pts	points
VRASPAC	Visual Reinforcement Assessment of the Perception of Speech Pattern Contrasts (test)
VRA	Visual Reinforcement Audiometry

Introduction

Current research on auditory development in children with profound sensorineural hearing loss emphasizes the critical importance of providing effective auditory stimulation as early as possible [1,2]. Current guidelines recommend that all infants should undergo hearing screening by 1 month of age. If the screening results are positive, a comprehensive audiological diagnosis should be completed by 3 months, followed by early intervention initiated by 6 months [3]. In infants with congenital profound sensorineural hearing loss, cochlear implantation (CI) is recommended, preferably within the first 12 months of life [4–6]. This timing is recognized as critical for achieving effective auditory stimulation of the auditory system. To ensure optimal outcomes following an early CI, it is crucial to assess and monitor the initial stages of auditory development. This necessitates the use of appropriate tools and methodologies tailored to evaluate and support auditory progress.

Auditory development

The development of the auditory pathway enables the auditory system to serve as the anatomical and physiological foundation for a wide range of perceptual abilities [7]. The classical theory is that the information processing in the auditory system proceeds hierarchically. Higher-order auditory areas make use of input from lower-order regions to perform increasingly complex operations. In this way, the highest cognitive functions integrate contextual cues and prior knowledge to generate meaningful auditory information.

Aslin and Smith [7] proposed a three-stage model of perceptual development, comprising: Level I – Sensory Primitives, which is basic sensory perception; Level II – Perceptual Representations, which entails complex neural coding at higher processing levels; and Level III – Higher-Order Representations, which encompasses cognitive and linguistic processing. Building on this model, Carney set out a linear pathway for auditory perceptual development in infants and toddlers [8]. The corresponding stages in auditory perception are then defined as: Level I – Sound Awareness, the ability to detect auditory stimuli; Level II – Sound Discrimination, reflecting the capacity to differentiate between one sound and another; and Level III – Sound Identification, involving the extraction of meaning from sound sources [9].

The maturation and functional integration of the structures of auditory pathway occur during the period when they start to be actively engaged in sound perception [1]. Synchronous activity of neurons within the pathway serves as a stimulus, facilitating the formation of connections and leading to more efficient auditory processing. The changes in neural connections that occur during development as a result of environmental interactions are referred to as developmental neuroplasticity, while the timeframe during which the brain is particularly receptive to these changes is known as the critical period [10]. Within the critical period, even brief exposure to stimuli can significantly affect the final organization and function of the neuronal network.

In summary, auditory perception depends on the successful structural and functional maturation of the auditory pathway, especially the development of later neural processing stages. The problem in children with congenital profound sensorineural hearing loss is that the absence of acoustic stimuli leads to a lack of neuronal activity in the auditory pathway [11–14]. The lack of neural activity leads to a weakening, or even loss, of synaptic connections, and this can cause permanent defects in the central auditory pathway [15].

Neural consequences of congenital profound sensorineural hearing loss

The three primary consequences of congenital profound sensorineural hearing loss are:

1. Intramodal deficits. The absence of auditory stimulation during critical developmental periods leads to the underdevelopment and dysfunction of auditory neural circuits, impairing their ability to accurately process sensory information [16–18].
2. Cross-modal plasticity. The sensory deprivation in the auditory system results in compensatory recruitment by other sensory modalities, a phenomenon that may disrupt neurosensory restoration efforts during later stages of development [19,20].
3. Reduced multimodal information processing capacity. The limitations in auditory input not only affect hearing-related pathways but also constrain the integration and processing of information across multiple sensory domains [21].

The observed deficits indicate that the auditory cortex in congenital profound sensorineural hearing loss, while capable of basic stimulus detection, lacks the robust feature representation necessary for complex auditory discrimination and object identification [19]. In children with congenital profound sensorineural hearing loss who receive cochlear implants (CIs) at a later age, the auditory system exhibits functional optimization for basic stimulus detection (Level I in the Aslin and Smith model) rather than for processing and discriminating fine auditory details (Levels II and III in the model).

CIs for children with congenital profound sensorineural hearing loss

Cochlear implants have been confirmed to be effective in providing auditory input to the auditory system. Numerous studies have demonstrated that in children with congenital profound sensorineural hearing loss, a CI allows for development of auditory perception milestones, including a child's ability to understand speech [12,22–24]. It has also been shown that the use of CIs in children with congenital profound sensorineural hearing loss is far more effective than the use of hearing aids [25–28]. Additionally, the age at implantation has been found to be a determining factor in auditory development [23,29,30].

The optimal age for receiving a CI in children with congenital profound sensorineural hearing loss has been investigated using cortical auditory evoked potentials (CAEPs), in particular the latency of the first positive peak (P1) of the waveform [31,32]. The latency of P1 reflects the efficiency of peripheral and central auditory processing. In normally developing children it shortens with age and is therefore considered a biomarker for auditory system maturation [31].

The results of such studies support the existence of a critical period for auditory development. In children with congenital profound sensorineural hearing loss who received CIs before the age of 3.5, their P1 latencies followed the same trajectory as normal hearing children [32,33].

However, in children who received CIs after the age of 7 years, the P1 latencies were significantly prolonged and did not change with the duration of CI experience. This strongly suggests that exposure to sound early in life is essential for appropriate auditory development. These electrophysiological findings are consistent with behavioral studies, which show that children who receive CIs before the age of 2 exhibit faster and more age-appropriate speech and language development than those who were implanted later [4,12,23,34].

At the same time, it has been confirmed that CI surgery is safe for children less than 1 year old [5,35]. Indeed, research shows that CIs are most effective in this group of children [4–6,25,29,30,36,37]. However, when a child with congenital profound sensorineural hearing loss receives a CI during their critical period of auditory development, it is important to evaluate its efficacy. A major difficulty in evaluating a young child is selecting an age-appropriate tool that allows their auditory development to be continually monitored from intervention onwards.

Tools for monitoring auditory development

Auditory development is usually assessed with two types of assessment methods, behavioral and electrophysiological. Behavioral tests, such as Visual Reinforcement Audiometry (VRA), used from 5 months of age, or Conditioned Play Audiometry (CPA) for children older than 2 years [38], only allow the ability to detect sound (Level I) to be assessed. However, tests are being developed to evaluate other stages of auditory development. The Visual Reinforcement Assessment of the Perception of Speech Pattern Contrasts test (VRASPAC) can assess the discrimination stage of auditory development (Level II) in children as young as 9 months [39]. Nevertheless, the majority of tests are only suitable for evaluating speech identification (Level III) in children older than 3 years; examples are the Automated McCormick Toy Discrimination Test [40], the Adaptive Auditory Speech Test (AAST) [41], the Northwestern University–Children's Perception of Speech test (NU-CHIPS) [42], the Early Speech Perception (ESP) test, and the Pediatric Speech Intelligibility (PSI) test [43].

Behavioral assessments can be limited by the child's state during testing, such as their attentiveness, motor skills to perform the response task (e.g., head turning, manipulation of objects, picture pointing, button pushing), as well as by their receptive and expressive language skills. The tests require high clinician expertise and careful interpretation of the child's behavior. The second audiological method for evaluating early auditory development is to use electrophysiological measures. However, it is difficult to directly translate the amplitude or latency of acoustically evoked auditory potentials into detection, discrimination, or identification ability [33].

Auditory development can also be evaluated based on criteria-referenced rating scales such as the Ling Developmental Scales [44], the Listening Progress Profile (LIP), and the Meaningful Auditory Integration Scale (MAIS) [45]. These assessments share a similar limitation as behavioral ones: their evaluation relies on the patient's attentiveness and the clinician's involvement.

Another possible avenue is to use parental questionnaires which ask questions about the child's auditory behavior in everyday situations. Questionnaire items gauge the child's reactions to a variety of environmental sounds and voices, their ability to discriminate between sounds, and their skill in deriving meaning from them.

Parental questionnaires

The most widely used parental questionnaires include the Auditory Behavior in Everyday Life (ABEL) [46]; the Children's Home Inventory for Listening Difficulties (CHILD) [47]; Early Listening Function (ELF) [48]; Functional Auditory Performance Indicators (FAPI) [49]; LittlEARS Auditory Questionnaire (LEAQ) [50]; Parent's Evaluation of Aural/Oral Performance of Children (PEACH) [51]; and the Infant–Toddler Meaningful Auditory Integration Scale (IT-MAIS) [52].

Numerous studies have confirmed that such questionnaires can provide reliable information about the child's development [53–55]. However to be diagnostically acceptable, a questionnaire needs to have been prepared on the basis of a theoretical framework, be validated, and have psychometric properties suitable for measuring the defined construct [56,57]. Suitable criteria for assessment tools have been proposed by Andresen [58] and include characteristics related to conceptual clarity (covering the relevant construct intended to be measured), the availability of norms/ standard values, explanation of the measurement model, description of item/instrument bias, respondent and administrative burden, reliability, discriminant validity (ability to differentiate subgroups that are expected to differ), convergent validity (validated against a gold standard and/or with confirmed good psychometric properties), ecological validity (evaluates the subject in a realistic environment), responsiveness (sensitivity to important changes in interventions), culture/language adaptations, and alternate/accessible forms.

In their *Critical Review of Audiological Outcome Measures for Infants and Children*, Bagatto and colleagues [59] reviewed parental questionnaires according to criteria proposed by Andresen [58] and rated them using a three-point scale (A, B, C). For the other above-mentioned questionnaires, data on reliability and validity were provided for ABEL, CHILD, IT-MAIS, LEAQ, and PEACH. Only for IT-MAIS, LEAQ, and PEACH were normative data available; only for LEAQ and PEACH was there a description of a measurement model; and only for LEAQ and PEACH were versions available in languages other than the original. Overall, LEAQ received the highest rating among the evaluated questionnaires. However, Bagatto and colleagues highlight a limitation in the LEAQ questionnaire: the lack of an assessment of its responsiveness. There was insufficient evidence to determine whether LEAQ scores are sensitive to significant changes following interventions [58].

Material and methods

LittlEARS Auditory Questionnaire

The LEAQ questionnaire has gained recognition among researchers and clinicians due to its clearly described measurement model, evidence of its validity, availability of normative data, and low respondent and administrative burden. This has resulted in an increasing number of linguistic adaptations and ongoing research [60–65]. Studies using different linguistic versions provide further evidence of the questionnaire's responsiveness and sensitivity to major interventional changes [64,66,67]. Obrycka et al. [30] collected evidence for discriminant validity. The authors identified differences in LEAQ scores among the following groups: 1) children implanted before 12 months of age compared to those implanted between 12 and 24 months; 2) children with extended versus shorter experience with hearing aids prior to CI; and 3) children who, before implantation, exhibited responses across a wide frequency range with their hearing aids compared to children who did not [30]. Wang et al. [68] found a negative correlation between LEAQ total score and P1 latency of CAEPs, with higher scores corresponding to shorter P1 latency (a biomarker for auditory system maturation). This provides further evidence that LEAQ measures auditory development and demonstrates convergent validity.

LEAQ was developed to assess the auditory development of infants and toddlers following a CI by measuring hearing abilities as they get older [50]. It consists of 35 questions for parents, organized according to the milestones of auditory development that children typically acquire over the first 24 months of life. The results of the questionnaire fall in the range 0 to 35 points depending on the child's age, although the expected (mean) value for normally developing children varies from approximately 3 pts in newborns to 33 pts in 2-year-olds (green lines in **Figures 1a, 1b**).

The structure of LEAQ is organized such that the 35 items increase in difficulty in line with the Aslin and Smith three-level model of auditory development: Level I (detection), Level II (discrimination), and Level III (identification). Items 1–16 focus primarily on Levels I and II (detection and discrimination) and assess the child's responses to human voices, music, environmental sounds, and sound-producing toys. Items 17–35, which relate to Level III of auditory development, evaluate the child's ability to identify sounds by, for example, associating names with objects or following spoken instructions.

The results of LEAQ can only be sensibly interpreted in terms of the child's age. For example, in **Figure 1a** the blue point is placed at the intersection of the horizontal line representing the child's LEAQ total score (15 points) and the vertical line representing the child's age (10 months). Any result above the minimum value (red dashed line) is interpreted as age-appropriate. A result falling on the green line reflects a value expected from an average normal hearing (NH) child (**Figure 1b**). Such a visualization is helpful in everyday clinical practice, when the trajectory of auditory development in a child can be analyzed and discussed with the parents.

Despite its numerous advantages, the LEAQ questionnaire is not without limitations. The primary limitation of the LEAQ questionnaire is its restricted period of applicability, being validated only for children up to 2 years of age [60,61]. From a clinical standpoint, there is a need to facilitate longitudinal assessment of auditory development
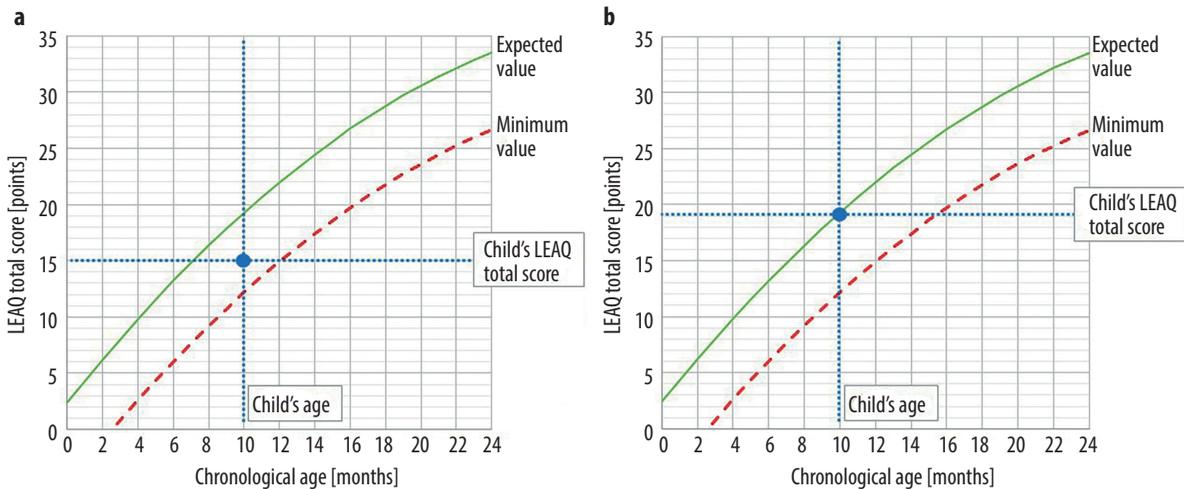
Figure 1. Interpretations of LEAQ total scores. **a**) Total score (blue dot) above minimum value (red dashed line), indicating satisfactory auditory development for a 10-month-old with a CI. **b**) Total score of 10-month-old equal to the average score of a normal hearing 10-month-old child (blue dot coinciding with green line of expected values)

beyond this age using the same tool. Consequently, the 2-year age limit represents a significant constraint.

Additionally, the LEAQ should not be employed for children older than 24 months, as the absence of normative data for this age group renders interpretation of results problematic. This limitation is particularly relevant in cases where intervention is delayed, and the child exceeds 2 years of age. In such cases, interpretations derived from the LEAQ should be approached with caution due to the saturation of normative data for older children. The following sections address this issue and other challenges associated with interpreting results, particularly those involving group analyses.

Challenges in interpreting LEAQ scores

The challenge comes when analyzing data from a cohort of children, for example when trying to assess the effectiveness of a CI over time or performing between-group comparisons. The traditional approach of comparing mean results before a CI and 1 year afterwards can produce misleading conclusions.

Firstly, as already said, the LEAQ total score is meaningless without reference to the child's age. Thus, a mean total score of 8 pts might be expected for a 3-month-old baby, be a minimum for a 7-month-old, and be far below the minimum for an 18-month-old (points A, B, and C in **Figure 2**).

Secondly, the age at which children receive a CI varies considerably. For example, Liu and colleagues [69] evaluated 33 children implanted between 6 and 46 months of age. The average LEAQ total scores for the group at 1, 12, and 24 months of CI use were 5, 24, and 33 pts respectively. When analyzing scores at 1 month post CI, for the youngest children (7 months of age) this score is very close to the normative range (2 pts below the minimum score), whereas for children receiving a CI later, the distance away from the normative range grows progressively larger (around 22
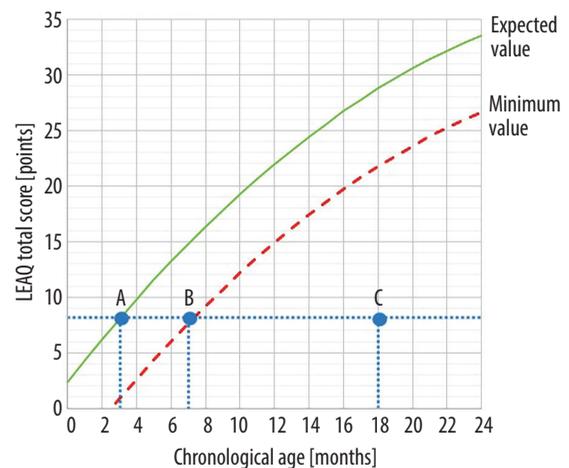

Figure 2. By itself, an LEAQ score is meaningless without knowing the child's age. For example, an LEAQ score of 8 points could come from a normal baby 3 months old (A), from a 7-month-old baby at minimum value (B), or from an 18-month-old far below the minimum (C)

pts for children at 24 months) until it becomes undefined for children older than 24 months (**Figure 3**, blue arrows).

Looking now at scores 12 months post CI, for the youngest children (6 months old at CI, 18 months old after 1 year of CI use), a result of 24 points indicates age-appropriate auditory development (5 pts below the expected score, but 2 pts above the minimum value). On the other hand, for the oldest children (46 months at CI and 58 months after 12 months of CI use) a score of 24 points indicates a substantial gain in auditory development compared to the initial score, but nevertheless it cannot be compared to NH children due to the age range substantially exceeding 24 months (**Figure 3**, orange arrows). To properly assess these children, a different age-appropriate tool is needed.
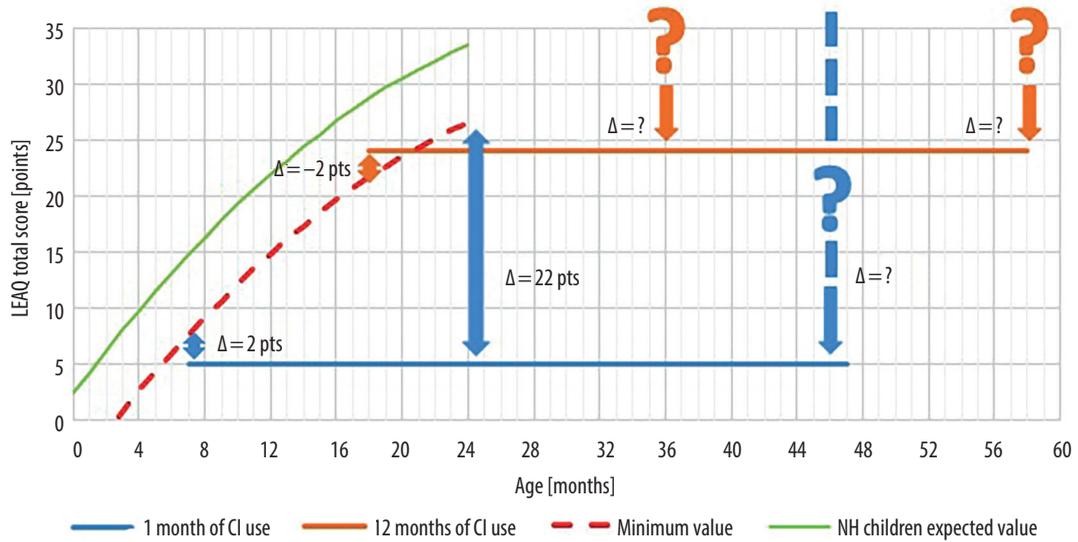
Figure 3. Relation between normative curve and average LEAQ total scores in a group of children implanted at ages ranging from 6 to 46 months. Blue line: after 1 month of CI use; orange line: after 12 months of CI use
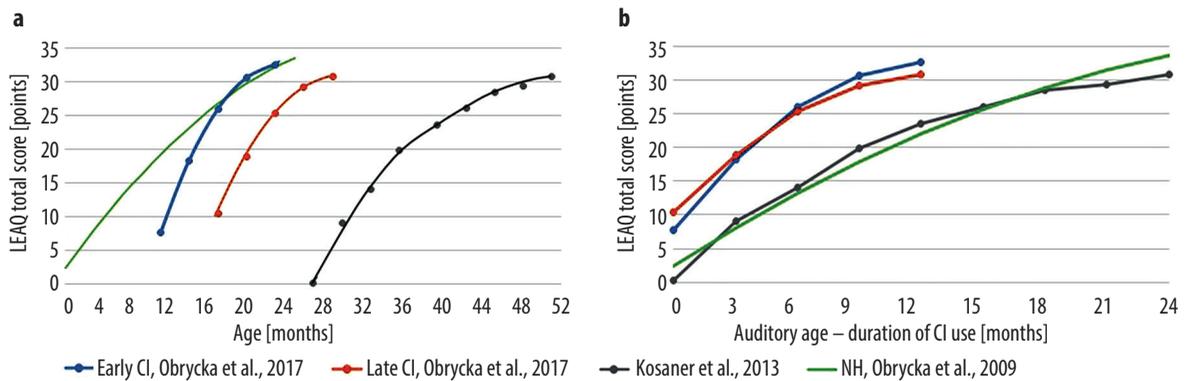
Figure 4. Two ways of plotting LEAQ total scores for groups of children implanted "early" (before 12 months of age, blue lines), "late" (12–24 months, orange lines), and "out-of-LEAQ-range" (black lines for data from Kosaner et al. 2013 [66]). **a**) Data plotted as a function of chronological age; **b**) data plotted as a function of auditory age (duration of CI use). Green lines are data for normal hearing children which extends to 24 months

We can therefore see that comparing means only makes sense within a group of children implanted at similar ages and later assessed at a time within the range where LEAQ remains applicable (i.e. less than 2 years of age). Such an analysis was performed by Honigman and colleagues when they used LEAQ to assess CI effectiveness in 134 children implanted within the age range of 9 to 11 months [70]. LEAQ was administered pre-operatively and post-operatively out to 2 years. A comparison of pre-implant score (mean 5.2, *SD* 6.3) to post-implant score (mean 27.3, *SD* 7.5) indicates substantial progress in auditory development. Nevertheless, such a comparison requires additional exploration into whether the results are age-appropriate. A total score of 5.2 pts for children at age 9–11 months is far below the minimum value (which ranges from 10.7 to 13.6 pts). A score of 27.3 pts after 2 years of CI use cannot be interpreted in the same way (using normative values) since the age of the tested children is now 33–35 months, which is beyond the LEAQ assessment range. A

good example of such an "out of range" assessment is presented in **Figure 4a**.

To overcome the limitation of going beyond the standard age range for LEAQ assessment, a number of studies have presented their results in terms of auditory age. For children with normal hearing, auditory age is of course the same as chronological age. But for implanted children, it is assumed that auditory development only begins when the implant is activated, so that auditory age then becomes the duration of CI use. The results are plotted in a similar way except that the *x*-axis is labelled as auditory age (or hearing age, or time after CI switch-on, etc.) [64,66,67,71,72]. For example, Yidi and colleagues [71] tested 287 children implanted at ages of 7 to 36 months. Taking the individual results, the authors built a model of auditory development from CI activation to 20 months of CI use. The authors concluded that auditory development in CI children was approximately the same as in NH children [71].

But the concern here is that, after 24 months of CI use, the older children were 5 years old but only had the level of auditory development of a NH child 2 years old. This represents a substantial developmental delay, since at age 5 much more complex auditory ability is expected (Level III, object identification) but it is beyond the range probed by LEAQ.

As an aside, it is not uncommon in LEAQ studies that the trajectory of total score as a function of duration of CI use falls above the normative curve. Keep in mind, however, that such graphs are usually overviews of early auditory development in CI children who are 2–3 years older than normal hearing children [66,67,71] (and sometimes even 5 years older [64]). Such graphs do not show the true relationship between auditory development in NH and CI children. The real goal of implantation is actually to reduce the delay, and examples of how the gap can be reduced are presented in **Figure 4b**.

Another complication is the assumption that auditory age should be counted from CI surgery or activation. In fact, there is a huge variability in LEAQ scores at this time. Scores may range from 0 to 17 pts [71], 20 pts [67], or even up to 33 pts [64]. This suggests that many children achieve some level of auditory development before receiving a CI and so their auditory age should not necessarily be counted from CI activation.

Calculation of auditory development delay

The huge variability in preoperative scores can be taken into account by using normative values for auditory age calculations [25,27]. For example, if the LEAQ total score is 20 points (**Figure 5**) we look for the age at which a child with normal hearing achieves 20 points (by moving left along the *x*-axis to the green line). For the Polish version of LEAQ this occurs at 10.5 months of age. Thus, if a child's LEAQ total score is 20 points, we determine that, based on expected values, the child's auditory age is 10.5 months, irrespective of actual age. Then, knowing the child's chronological age, we calculate the *delay* in auditory development as the difference between the chronological age and the auditory age. For example, if a 20-month-old child has a total score of 20 pts then its auditory age is 10.5 months and the delay in auditory development is 9.5 months. That is, the auditory age corresponds to the age at which the horizontal line intersects the normative curve, and the delay is the horizontal gap between the actual LEAQ total score and the norm.

Calculating delays in auditory development in this way have been used in some studies on the effectiveness of CIs based on the PICO (patient, intervention, comparator, outcome) schema [25,27]. In this work, 32 children (P) implanted before the age of 12 months were compared (C) to 19 children who used hearing aids. Both groups of children were fitted with their respective devices at approximately the same age; they were also matched for age and degree of hearing loss. Some 10 months after the initial fitting of their devices, the children were assessed using LEAQ. The LEAQ total score was used to determine the delay (O) in auditory development. On average, the auditory development delay in the hearing aid group was
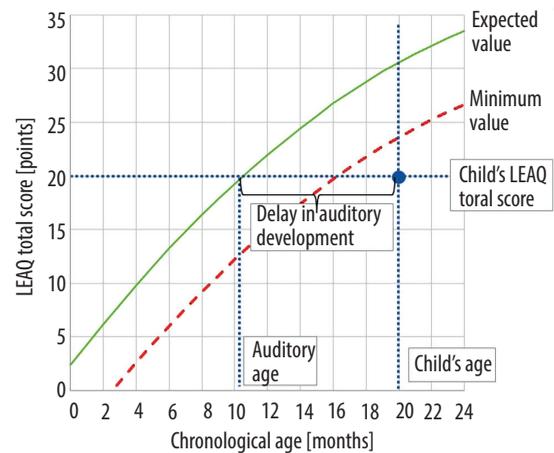
Figure 5. Interpreting LEAQ scores in terms of delay in auditory development. The example is a 20-month-old child with an LEAQ total score of 20 pts (blue dot). This same score would be achieved by a normal child at 10.5 months, meaning that in this case the auditory age is 10.5 months and the delay in auditory development is 9.5 months

14.3 months longer than in the CI group after 10 months of device use.

Although calculation of the delay in auditory development allows for the assessment of CI effectiveness in relation to normal hearing children, this method still has some limitations. The 9.5 months delay in auditory development, as presented in **Figure 5**, may seem to be insignificant. However, if we take into account a child's age, then for a 20-month-old child a 9.5-month delay means 48% of their life, while for a 9.5-month-old child, a 9.5-month delay corresponds to 100% of their life (there has been a complete lack of auditory development). Due to these difficulties in comparing the results of children implanted at different ages and at different stages of auditory development, a unified way to calculating auditory delay, one that takes into account a child's age, is proposed.

Relative delay in auditory development

The relative delay in auditory development is defined as the child's delay in auditory development divided by their chronological age. That is,

$$Relative\ delay = \frac{Delay\ in\ auditory\ development}{Chronological\ age} \times 100\%$$

$$= \frac{Chronological\ age - Auditory\ age}{Chronological\ age} \times 100\%$$

As described in the previous section, the auditory age can be derived from the child's LEAQ total score and a chart containing normative curves. A more accurate method is to calculate the hearing age from the normative curve equation. This requires solving a quadratic equation of the form $ax^2 + bx + c - y = 0$, where x is auditory age; y is the LEAQ

Table 1. Equations for calculating relative delay from different linguistic adaptations of LEAQ

Adaptation	Regression equation $(y = ax^2 + bx + c)$	Relative delay (D_{rel})
OVERALL [60]	$y = -0.038\,x^2 + 2.163\,x + 3.47$	$\dfrac{A_{chr} - \dfrac{-2.163 + \sqrt{4.679 + 0.152\,(3.47 - LEAQ)}}{-0.076}}{A_{chr}} \times 100\%$
Bulgaria [60]	$y = -0.018\,x^2 + 1.604\,x + 5.56$	$\dfrac{A_{chr} - \dfrac{-2.604 + \sqrt{2.573 + 0.072\,(5.56 - LEAQ)}}{-0.036}}{A_{chr}} \times 100\%$
Belgium [60]	$y = -0.016\,x^2 + 1.666\,x + 3.673$	$\dfrac{A_{chr} - \dfrac{-1.666 + \sqrt{2.776 + 0.064\,(3.376 - LEAQ)}}{-0.032}}{A_{chr}} \times 100\%$
Slovakia [60]	$y = -0.033\,x^2 + 2.147\,x + 4.143$	$\dfrac{A_{chr} - \dfrac{-2.147 + \sqrt{4.61 + 0.132\,(4.143 - LEAQ)}}{-0.066}}{A_{chr}} \times 100\%$
USA (English) [60]	$y = -0.047\,x^2 + 2.402\,x + 2.308$	$\dfrac{A_{chr} - \dfrac{-2.402 + \sqrt{5.77 + 0.188\,(2.308 - LEAQ)}}{-0.094}}{A_{chr}} \times 100\%$
Romania [60]	$y = -0.031\,x^2 + 1.919\,x + 2.538$	$\dfrac{A_{chr} - \dfrac{-1.919 + \sqrt{3.683 + 0.124\,(2.538 - LEAQ)}}{-0.062}}{A_{chr}} \times 100\%$
France [60]	$y = -0.049\,x^2 + 2.461\,x + 0.879$	$\dfrac{A_{chr} - \dfrac{-2.461 + \sqrt{6.507 + 0.196\,(0.879 - LEAQ)}}{-0.098}}{A_{chr}} \times 100\%$
Serbia [60]	$y = -0.046\,x^2 + 2.463\,x + 0.879$	$\dfrac{A_{chr} - \dfrac{-2.463 + \sqrt{6.066 + 0.184\,(0.879 - LEAQ)}}{-0.092}}{A_{chr}} \times 100\%$
Finland [60]	$y = -0.029\,x^2 + 1.947\,x + 4.586$	$\dfrac{A_{chr} - \dfrac{-1.947 + \sqrt{3.791 + 0.116\,(4.586 - LEAQ)}}{-0.058}}{A_{chr}} \times 100\%$
Slovenia [60]	$y = -0.033\,x^2 + 2.075\,x + 3.762$	$\dfrac{A_{chr} - \dfrac{-2.075 + \sqrt{4.306 + 0.132\,(3.762 - LEAQ)}}{-0.066}}{A_{chr}} \times 100\%$
Germany [60]	$y = -0.038\,x^2 + 2.217\,x + 2.066$	$\dfrac{A_{chr} - \dfrac{-2.217 + \sqrt{4.915 + 0.152\,(2.066 - LEAQ)}}{-0.076}}{A_{chr}} \times 100\%$
Russia [60]	$y = -0.072\,x^2 + 3.156\,x - 2.354$	$\dfrac{A_{chr} - \dfrac{-3.156 + \sqrt{9.96 + 0.288\,(-2.354 - LEAQ)}}{-0.144}}{A_{chr}} \times 100\%$
China [60]	$y = -0.038\,x^2 + 2.23\,x + 1.211$	$\dfrac{A_{chr} - \dfrac{-2.23 + \sqrt{4.973 + 0.152\,(1.211 - LEAQ)}}{-0.076}}{A_{chr}} \times 100\%$
USA (Spanish) [60]	$y = -0.026\,x^2 + 1.779\,x + 9.084$	$\dfrac{A_{chr} - \dfrac{-1.779 + \sqrt{3.165 + 0.104\,(9.084 - LEAQ)}}{-0.052}}{A_{chr}} \times 100\%$
Switzerland [60]	$y = -0.030\,x^2 + 1.921\,x + 4.687$	$\dfrac{A_{chr} - \dfrac{-1.921 + \sqrt{3.69 + 0.12\,(4.687 - LEAQ)}}{-0.06}}{A_{chr}} \times 100\%$

Table 1 continued. Equations for calculating relative delay from different linguistic adaptations of LEAQ

Adaptation	Regression equation $(y = ax^2 + bx + c)$	Relative delay (D_{rel})
Poland [61]	$y = -0.028\,x^2 + 1.969\,x + 2.396$	$\dfrac{A_{chr} - \dfrac{-1.952 + \sqrt{3.81 + 0.112\,(2.917 - LEAQ)}}{-0.056}}{A_{chr}} \times 100\%$
Greece [60]	$y = -0.064\,x^2 + 2.653\,x + 6.272$	$\dfrac{A_{chr} - \dfrac{-2.653 + \sqrt{7.038 + 0.256\,(6.272 - LEAQ)}}{-0.128}}{A_{chr}} \times 100\%$
Israel (Hebrew) [64]	$y = -0.036\,x^2 + 2.181\,x + 2.531$	$\dfrac{A_{chr} - \dfrac{-2.181 + \sqrt{4.757 + 0.152\,(2.531 - LEAQ)}}{-0.076}}{A_{chr}} \times 100\%$
Israel (Arabic) [64]	$y = -0.029\,x^2 + 1.862\,x + 4.744$	$\dfrac{A_{chr} - \dfrac{-1.862 + \sqrt{3.467 + 0.144\,(4.744 - LEAQ)}}{-0.072}}{A_{chr}} \times 100\%$
Canada (English) [62]	$y = -0.013\,x^2 + 1.55\,x + 6.55$	$\dfrac{A_{chr} - \dfrac{-1.55 + \sqrt{2.403 + 0.052\,(6.55 - LEAQ)}}{-0.026}}{A_{chr}} \times 100\%$
Spain [63]	$y = -0.052\,x^2 + 2.69\,x - 0.72$	$\dfrac{A_{chr} - \dfrac{-2.69 + \sqrt{7.236 + 0.208\,(-0.72 - LEAQ)}}{-0.104}}{A_{chr}} \times 100\%$
Ghana [65]	$y = -0.081\,x^2 + 3.303\,x + 0.648$	$\dfrac{A_{chr} - \dfrac{-3.303 + \sqrt{10.91 + 0.324\,(0.648 - LEAQ)}}{-0.162}}{A_{chr}} \times 100\%$

Note: A_{chr}, chronological age; *LEAQ*, LEAQ total score

total score, and *a*, *b*, *c* are coefficients of the quadratic function of auditory development determined for the particular language version of LEAQ used. Strictly, this equation has two solutions, but only one is in the range assessed with the questionnaire (0–24 months), so the auditory age can be specified as:

$$Auditory\ age = \frac{-b + \sqrt{b^2 - 4a\,(c - LEAQ\ score)}}{2a}$$

and relative delay of auditory development as:

$$Relative\ delay = \frac{Chronological\ age - \dfrac{-b + \sqrt{b^2 - 4a\,(c - LEAQ\ score)}}{2a}}{Chronological\ age} \times 100\%$$

The normative curve established from 16 studies by Coninx and colleagues is described by the equation $y = -0.038\,x^2 + 2.163\,x + 3.470$ [60]. Based on this curve the relative delay can be calculated as:

$$Relative\ delay = \frac{Chronological\ age - \dfrac{-2.163 + \sqrt{4.679 + 0.152\,(3.47 - LEAQ\ score)}}{-0.076}}{Chronological\ age} \times 100\%$$

For different linguistic adaptations, the corresponding equations for relative delay are presented in **Table 1**.

At this point, the proposed method was used to reanalyze data from two already published studies [30,66]. To calculate mean relative delay in the study by Obrycka et al. [30] the LEAQ results of 54 children implanted 'early' (before the age of 12 months, first group) and those of 68 children implanted 'late' (between 12 and 24 months, second group) were used. Relative delay was calculated based on the equation for Poland from Obrycka et al. (see **Table 1**). For the other study, Kosaner et al. [66], the results of 20 children implanted between 15 and 35 months (third group) were recalculated based on the 'overall' equation (top of **Table 1**). The mean relative delays for each group are shown in **Figure 6**.

Discussion

Based on current knowledge of the neural consequences of congenital profound sensorineural hearing loss in children and existing guidelines for early detection and intervention, such children should be provided with a CIs as early as possible [2–4,6,12,23,32]. Here, assessing early auditory development is of highest importance.

The LittlEARS Auditory Questionnaire (LEAQ) is an appropriate tool for monitoring early auditory development from the moment a CI is provided. However, some challenges in interpreting results from cohorts of CI children have been identified, primarily related to the variability in outcomes and the age at which children received
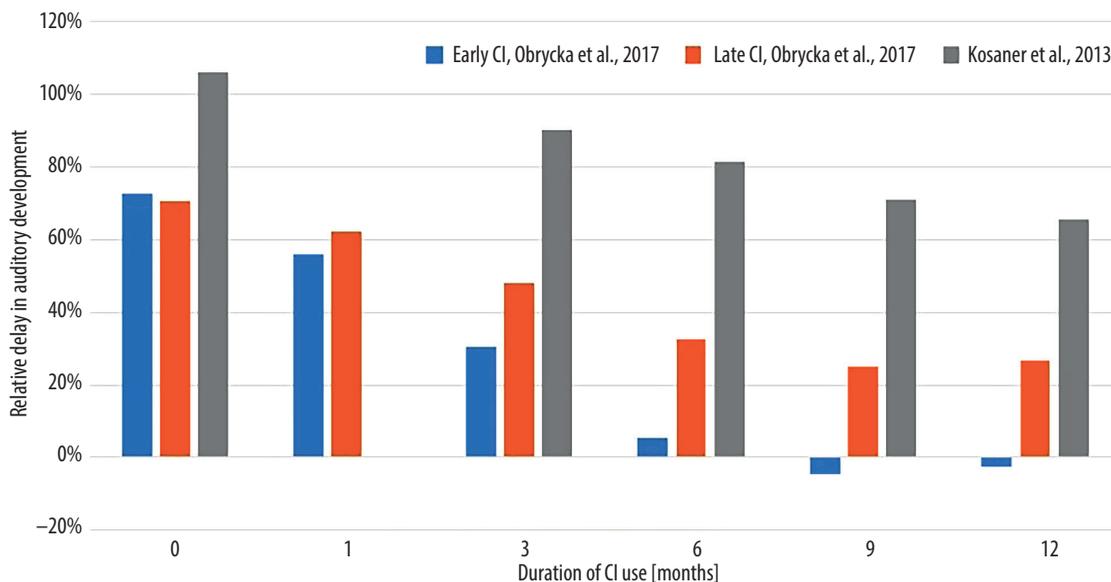
Figure 6. Relative delay in auditory development recalculated from studies by Obrycka et al. [30] and Kosaner et al. 2013 [66]

the intervention. To address these challenges, a method for calculating the *relative delay* in auditory development has been proposed. This method was applied to reanalyze data from three groups: children implanted "early" (before 12 months of age, first group), children implanted "late" (between 12 and 24 months of age, second group) as reported by Obrycka et al. [30], and a third group described by Kosaner et al [66].

In both groups examined from Obrycka et al. [30], relative delay at CI activation was lower (73% and 70% for early and late groups respectively) than in the group studied by Kosaner et al. [66] (110%). This indicates that the initial stage of auditory development (in terms of LEAQ total score) was higher in the Obrycka et al. study (7.7 pts in the early group and 10.5 pts in the late group) compared to Kosaner et al. (0.3 pts) (see **Figure 4a**). Moreover, the delay in auditory development in terms of distance from the normative curve is longer in the late group than in the early group (**Figure 4a**). However, the combination of longer delay in auditory development and older age in the late group yields similar relative delay to the early group who had shorter delays and younger ages. Additionally, the relative delay above 100% in the group evaluated by Kosaner et al. (**Figure 6**) comes from initial scores close to 0 pts, while in NH children the result at birth is around 3 pts, indicating that auditory development starts in the mother's womb (**Figure 2**).

Figure 6 shows that in all three groups there is a reduction in relative delay over time. The most effective relative delay reduction is observed in the early group who received their CIs at an average age of 10 months. In this group the relative delay after 9 months of CI use was −5% and at 12 months −3%. Negative values of relative delay indicate LEAQ total scores above the average for NH children. In the group implanted late (average age at CI of 16 months), after a further 12 months of CI use the relative delay was reduced to 27% and in the third group implanted

even later (mean age at CI of 26 months) the delay reached 66% over the same period.

Those differences are not clear when just mean LEAQ total score is analyzed (**Figure 4b**), especially between the groups implanted early and late. The recalculated results show a clear difference between the groups, whereas the traditional approach shows only a small difference after a year of using a CI. The proposed method therefore allows differences to be captured earlier on. Using the traditional approach, differences are small and can only be detected after an extended period of observation [30].

Finally, in all three groups the raw LEAQ plateaued, approaching maximum values. In the early group this indicated age-appropriate auditory development (relative delays even below 0%) whereas in the late group the maximum score still equated to a relative delay of 27%. The third group of CI children [66] approached maximum values about 2 years later than NH children, indicating a 62% relative delay. Such a large delay may hamper auditory processing and discrimination of fine auditory detail, equivalent to Levels II and III of the Aslin and Smith model. The large relative delay was due to late CI implantation, where the age at CI was significantly later than in the other two groups.

The proposed method of assessing relative delay appears to be more sensitive to between-group differences. Moreover, it has more precision in showing the degree of auditory development compared to previously used methods. This method may assist researchers and clinicians in reporting early auditory development in a manner that allows for comparisons across different languages, studies, and centers. However, an important issue arises: the LEAQ has been validated only for children older than 2 years. The use of this questionnaire in older children, such as when intervention is delayed or in children with comorbidities where behavioral assessment is challenging, may be helpful in monitoring changes in auditory development.

Nonetheless, the results cannot be compared to normative data, nor can they confirm that the child is developing faster than peers with normal hearing since the metrical age of the study group and the normative group are different. This finding has significant implications, since in children younger than 2 years, early auditory development (as evidenced by age-appropriate scores on the LEAQ) can serve as a reliable indicator of typical speech and language development. In contrast, such predictive validity is not observed in children older than 2 years.

Conclusions

To minimize the neurological consequences of congenital profound sensorineural hearing loss there is general consensus that a child should be provided with a CI early on, even before 12 months of age. When doing so, it is important to check that the intervention has been effective, i.e. it has allowed the child to reach all levels of early auditory development as described by the Aslin and Smith model. A good measure of CI effectiveness is the delay (in months) in the child's auditory development compared to NH children. If the delay can be eliminated during the first year of CI use, then it is quite possible that the child will attain age-appropriate hearing by 2 years of age. Such a degree of auditory development will form a solid foundation for the development of higher-order auditory capabilities.

An excellent tool for monitoring all levels of early auditory development, as well as its delay in infants and toddlers, is the LittlEARS Auditory Questionnaire (LEAQ). It can be used to assess the effectiveness of cochlear implants, beginning from the first moment of CI activation.

The method proposed here for assessing the relative delay in auditory development based on LEAQ eliminates two difficulties in interpreting results within a cohort of CI children: one is related to the variability of results and the second is the age at intervention. This unified method should facilitate between-group comparisons in single or multicenter studies, as well as improve meta-analysis of data in evidence-based practice.

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References

1. Eggermont JJ, Moore JK. Morphological and functional development of the auditory nervous system. In: Human Auditory Development. Werner LA, Fay RR, Popper AN, editors. Springer Handbook of Auditory Research 42. New York: Springer; 2012, 61–106.
https://doi.org/10.1007/978-1-4614-1421-6

2. Kral A. Auditory critical periods: a review from system's perspective. Neuroscience, 2013; 247: 117–33.
https://doi.org/10.1016/j.neuroscience.2013.05.021

3. Joint Committee on Infant Hearing. Year 2019 Position Statement: Principles and Guidelines for Early Hearing Detection and Intervention Programs. JEHDI, 2019; 4(2): 1–44. https://docslib.org/doc/2800007/principles-and-guidelines-for-early-hearing-detection-and-intervention-programs

4. Dettman SJ, Pinder D, Briggs RJS, Dowell RC, Leigh JR. Communication development in children who receive the cochlear implant younger than 12 months: risks versus benefits. Ear Hear, 2007; 28 (Suppl. 2): 11–8.
https://doi.org/10.1097/AUD.0b013e31803153f8

5. Colletti V, Carner M, Miorelli V, Guida M, Colletti L, Fiorino FG. Cochlear implantation at under 12 months: report on 10 patients. Laryngoscope, 2005; 115(3): 445–9.
https://doi.org/10.1097/01.mlg.0000157838.61497.e7

6. May-Mederake B. Early intervention and assessment of speech and language development in young children with cochlear implants. Int J Pediatr Otorhinolaryngol, 2012; 76(7): 939–46.
https://doi.org/10.1016/j.ijporl.2012.02.051

7. Aslin RN, Smith LB. Perceptual development. Annu Rev Psychol, 1988; 39(1): 435–73.
https://doi.org/10.1146/annurev.ps.39.020188.002251

8. Carney A. Audition and the development of oral communication competency. In: Amplification for Children with Auditory Deficits. Bess HF, Gravel JS, Tharpe A, editors. Nashville, Tennessee: Wilkerson Center Press; 1996, 29–54.

9. Litovsky R. Development of the auditory system. Handb Clin Neurol, 2015; 129: 55–72.
https://doi.org/10.1016/B978-0-444-62630-1.00003-2

10. Cramer KS, Coffin AB, Fay RR, Popper AN, editors. Auditory Development and Plasticity. Cham: Springer International Publishing; 2017. https://doi.org/10.1007/978-3-319-21530-3

11. Kral A, Yusuf PA, Land R. Higher-order auditory areas in congenital deafness: top-down interactions and corticocortical decoupling. Hear Res, 2017; 343: 50–63.
https://doi.org/10.1016/j.heares.2016.08.017

12. Kral A, O'Donoghue GM. Profound deafness in childhood. N Engl J Med, 2010; 363(15): 1438–50.
https://doi.org/10.1056/NEJMra0911225

13. Sharma A, Dorman MF, Kral A. The influence of a sensitive period on central auditory development in children with unilateral and bilateral cochlear implants. Hear Res, 2005; 203(1–2): 134–43. https://doi.org/10.1016/j.heares.2004.12.010

14. Sharma A, Gilley PM, Dorman MF, Baldwin R. Deprivation-induced cortical reorganization in children with cochlear implants. Int J Audiol, 2007; 46(9): 494–9.
https://doi.org/10.1080/14992020701524836

15. Kral A, Hartmann R, Tillein J, Heid S, Klinke R. Congenital auditory deprivation reduces synaptic activity within the auditory cortex in a layer-specific manner. Cereb Cortex, 2000; 10: 714–26. https://doi.org/10.1093/cercor/10.7.714

16. Kral A, Tillein J, Heid S, Klinke R, Hartmann R. Cochlear implants: cortical plasticity in congenital deprivation. Prog Brain Res, 2006; 157: 283–313. https://doi.org/10.1016/s0079-6123(06)57018-9. PMID: 17167917

17. Raggio MW, Schreiner CE. Neuronal responses in cat primary auditory cortex to electrical cochlear stimulation. III. Activation patterns in short- and long-term deafness. J Neurophysiol, 1999; 82(6): 3506–26.
https://doi.org/10.1152/jn.1999.82.6.3506

18. Kral A, Tillein J, Heid S, Hartmann R, Klinke R. Postnatal cortical development in congenital auditory deprivation. Cereb Cortex, 2005; 15(5): 552–62. https://doi.org/10.1093/cercor/bhh156

19. Kral A, Dorman MF, Wilson BS. Neuronal development of hearing and language: cochlear implants and critical periods. Annu Rev Neurosci, 2019; 42: 47–65. https://doi.org/10.1146/annurev-neuro-080317-061513

20. Kral A, Sharma A. Crossmodal plasticity in hearing loss. Trends Neurosci, 2023; 46(5): 377–93. https://doi.org/10.1016/j.tins.2023.02.004

21. Kral A, Kronenberger WG, Pisoni DB, O'Donoghue GM. Neurocognitive factors in sensory restoration of early deafness: a connectome model. Lancet Neurol, 2016; 15(6): 610–21. https://doi.org/10.1016/S1474-4422(16)00034-X

22. Bond M, Mealing S, Anderson R, Elston J, Weiner G, Taylor RS, et al. The effectiveness and cost-effectiveness of cochlear implants for severe to profound deafness in children and adults: a systematic review and economic model. Health Technol Assess, 2009; 13(44). https://doi.org/10.3310/hta13440

23. Niparko JK, Tobey EA, Thal DJ, Eisenberg LS, Wang NY, Quittner AL, et al. Spoken language development in children following cochlear implantation. JAMA, 2010; 303(15): 11498–1506. https://doi.org/10.1001/jama.2010.451

24. Waltzman SB, Cohen NL, Gomolin RH, Shapiro WH, Ozdamar SR, Hoffman RA. Long-term results of early cochlear implantation in congenitally and prelingually deafened children. Am J Otol, 1994; 15 (Suppl 2): 9–13.

25. Lorens A, Obrycka A, Skarzynski H. Assessment of early auditory development in children after cochlear implantation. In: Advances in Audiology and Hearing Science: Volume 2. Hatzopoulos S, Ciorba A, Krumm M, editors. Apple Academic Press; 2020, 3–24. https://doi.org/10.1201/9780429292620-2

26. Mildner V, Šindija B, Zrinski KV. Speech perception of children with cochlear implants and children with traditional hearing aids. Clin Linguist Phon, 2006; 20(2–3): 219–29. https://doi.org/10.1080/02699200400027031

27. Obrycka A, Lorens A, Piotrowska A, Skarżyński H. [The LittlEARS questionnaire to assess the effectiveness of cochlear implantation in very young profoundly deaf children]. Now Audiofonol, 2014; 3(5): 52–8 [in Polish]. https://doi10.17431/893896

28. Tomblin JB, Spencer L, Flock S, Tyler R, Gantz B. A comparison of language achievement in children with cochlear implants and children using hearing aids. J Speech Lang Hear Res, 1999; 42(2): 497–511. https://doi.org/10.1044/jslhr.4202.497

29. Govaerts PJ, De Beukelaer C, Daemers K, De Ceulaer G, Yperman M, Somers T, et al. Outcome of cochlear implantation at different ages from 0 to 6 years. Otol Neurotol, 2002; 23(6): 885–90. https://doi.org/10.1097/00129492-200211000-00013

30. Obrycka A, Lorens A, Padilla García J-L, Piotrowska A, Skarzynski H. Validation of the LittlEARS Auditory Questionnaire in cochlear implanted infants and toddlers. Int J Pediatr Otorhinolaryngol, 2017; 93: 107–16. https://doi.org/10.1016/j.ijporl.2016.12.024

31. Sharma A, Dorman MF, Spahr AJ. Rapid development of cortical auditory evoked potentials after early cochlear implantation. Neuroreport, 2002; 13(10): 1365–8. https://doi.org/10.1097/00001756-200207190-00030

32. Sharma A, Dorman MF. Central auditory development in children with cochlear implants: clinical implications. Adv Otorhinolaryngol, 2006; 64: 66–88. https://doi.org/10.1159/000094646

33. Sharma A, Dorman MF, Spahr AJ. A sensitive period for the development of the central auditory system in children with cochlear implants: implications for age of implantation. Ear Hear, 2002; 23(6): 532–9. https://doi.org/10.1097/00003446-200212000-00004

34. Nicholas JG, Geers AE. Spoken language benefits of extending cochlear implant candidacy below 12 months of age. Otol Neurotol, 2013; 34(3): 532–8. https://doi.org/10.1097/MAO.0b013e318281e215

35. Valencia DM, Rimell FL, Friedman BJ, Oblander MR, Helmbrecht J. Cochlear implantation in infants less than 12 months of age. Int J Pediatr Otorhinolaryngol, 2008; 72(6): 767–73. https://doi.org/10.1016/j.ijporl.2008.02.009

36. Holman MA, Carlson ML, Driscoll CL, Grim KJ, Petersson RS, Sladen DP, et al. Cochlear implantation in children 12 months of age and younger. Otol Neurotol, 2013; 34(2): 251–8. https://doi.org/10.1097/MAO.0b013e31827d0922

37. Holt RF, Svirsky MA. An exploratory look at pediatric cochlear implantation: is earliest always best? Ear Hear, 2008; 29(4): 492–511. https://doi.org/10.1097/AUD.0b013e31816c409f

38. Cole EB, Flexer C. Children with Hearing Loss: Developing Listening and Talking, Birth to Six. First Edition. San Diego: Plural Publishing; 2007.

39. Eisenberg LS, Martinez AS, Boothroyd A. Assessing auditory capabilities in young children. Int J Pediatr Otorhinolaryngol, 2007; 71(9): 1339–50. https://doi.org/10.1016/j.ijporl.2007.05.017

40. Ousey J, Sheppard S, Twomey T, Palmer AR. The IHR-McCormick Automated Toy Discrimination test: description and initial evaluation. Br J Audiol, 1989; 23(3): 245–9. https://doi.org/10.3109/03005368909076506

41. Miggiani P, Coninx F, Schaefer K. Validation of the Maltese Adaptive Auditory Speech Test (AAST). Audiol Res, 2022; 12(4): 357–76. https://doi.org/10.3390/audiolres12040037

42. Elliott L, Katz D. Development of a new children's test of speech discrimination. St. Louis: Auditec; 1980.

43. Jerger S, Jerger J. Pediatric Speech Intelligibility Test. St. Louis: Auditec; 1984.

44. Ling AH. Schedules of development in audition, speech, language, communication for hearing-impaired infants and their parents. Washington, D.C.: Alexander Graham Bell Association for the Deaf; 1977.

45. Allum JHJ, Greisiger R, Straubhaar S, Carpenter MG. Auditory perception and speech identification in children with cochlear implants tested with the EARS protocol. Br J Audiol, 2000; 34(5): 293–303. https://doi.org/10.3109/03005364000000141

46. Purdy SC, Farrington DR, Moran CA, Chard LL, Hodgson SA. A parental questionnaire to evaluate children's auditory behavior in everyday life (ABEL). Am J Audiol, 2002; 11(2): 72–82. https://doi.org/10.1044/1059-0889(2002/010)

47. Anderson KL, Smaldino JJ. Children's home inventory of listening difficulties (CHILD). Available from: https://successforkidswithhearingloss.com/wp-content/uploads/2011/08/CHILD_pgs3-4.pdf [Accessed 24.1.2025]

48. Anderson K. ELF – Early Listening Function. Available from: https://successforkidswithhearingloss.com/wp-content/uploads/2011/08/ELF-Oticon-version.pdf [Accessed 24.1.2025]

49. Stredler-Brown A, Johnson D. Functional Auditory Performance Indicators (FAPI). Available from: https://www.phonakpro.com/content/dam/phonakpro/gc_hq/en/resources/counseling_tools/documents/child_hearing_assessment_functional_auditory_performance_indicators_fapi_2017.pdf [Accessed 24.1.2025]

50. Weichbold V, Tsiakpini L, Coninx F, D'Haese P. [Development of a parent questionnaire for assessment of auditory behaviour of infants up to two years of age]. Laryngo Rhino Otol, 2005; 84: 328–34 [in German].

51. Ching TYC, Hill M. The Parents' Evaluation of Aural/Oral Performance of Children (PEACH) scale: normative data. J Am Acad Audiol, 2007; 18(3): 220–35. https://doi.org/10.3766/jaaa.18.3.4

52. Zimmerman-Phillips S, Osberger MRA. Infant–Toddler Meaningful Auditory Integration Scale. Sylmar, CA: Advanced Bionics Corp.; 1997.

53. Archbold SM, Lutman ME, Gregory S, O'Neill C, Nikolopoulos TP. Parents and their deaf child: their perceptions three years after cochlear implantation. Deaf Educ Int, 2002; 4(1): 12–40. https://doi.org/10.1002/dei.114

54. Hoey H, McGee H, Fitzgerald M, Mortensen H, Hougaard P, Lynggaard H, et al. Parent and health professional perspectives in the management of adolescents with diabetes: development of assessment instruments for international studies. Qual Life Res, 2006; 15(6): 1033–42. https://doi.org/10.1007/s11136-006-0042-8

55. Watson LR, Baranek GT, Crais ER, Reznick SJ, Dykstra J, Perryman T. The First Year Inventory: retrospective parent responses to a questionnaire designed to identify one-year-olds at risk for autism. J Autism Dev Disord, 2007; 37(1): 49–61. https://doi.org/10.1007/s10803-006-0334-4

56. International Test Commission. International Guidelines for Test Use. Int J Test, 2001; 1(2): 93–114. https://doi.org/10.1207/s15327574ijt0102_1

57. Osterlind S. Constructing test items: multiple-choice, constructed-response, performance, and other formats. Boston: Kluwer Academic Publishers; 2001.

58. Andresen EM. Criteria for assessing the tools of disability outcomes research. Arch Phys Med Rehabil, 2000; 81 (12 Suppl 2): 15–20. https://doi.org/10.1053/apmr.2000.20619

59. Bagatto MP, Moodie ST, Seewald RC, Bartlett DJ. A critical review of audiological outcome measures for infants and children. Trends Amplif, 2011; 15(1): 23–33. https://doi.org/10.1177/1084713811412056

60. Coninx F, Weichbold V, Tsiakpini L, Autrique E, Bescond G, Tamas L, et al. Validation of the LittlEARS® Auditory Questionnaire in children with normal hearing. Int J Pediatr Otorhinolaryngol, 2009; 73(12): 1761–8. https://doi.org/10.1016/j.ijporl.2009.09.036

61. Obrycka A, Padilla García JL, Pankowska A, Lorens A, Skarzynski H. Production and evaluation of a Polish version of the LittlEars questionnaire for the assessment of auditory development in infants. Int J Pediatr Otorhinolaryngol, 2009; 73(7): 1035–42. https://doi.org/10.1016/j.ijporl.2009.04.010

62. Bagatto MP, Brown CL, Moodie ST, Scollie SD. External validation of the LittlEARS® Auditory Questionnaire with English-speaking families of Canadian children with normal hearing. Int J Pediatr Otorhinolaryngol, 2011; 75(6): 815–7. https://doi.org/10.1016/j.ijporl.2011.03.014

63. García Negro AS, Padilla García JL, Sainz Quevedo M. Production and evaluation of a Spanish version of the LittlEARS® Auditory Questionnaire for the assessment of auditory development in children. Int J Pediatr Otorhinolaryngol, 2016; 83: 99–103. https://doi.org/10.1016/j.ijporl.2016.01.021

64. Geal-Dor M, Jbarah R, Meilijson S, Adelman C, Levi H. The Hebrew and the Arabic version of the LittlEARS® Auditory Questionnaire for the assessment of auditory development: results in normal hearing children and children with cochlear implants. Int J Pediatr Otorhinolaryngol, 2011; 75(10): 1327–32. https://doi.org/10.1016/j.ijporl.2011.07.030

65. Kayode O, Adeyemo AA. The Yoruba version of LittlEARS Auditory Questionnaire: Evaluation of auditory development in children with normal hearing. J Otol, 2018; 13(3): 92–6. https://doi.org/10.1016/j.joto.2018.07.001

66. Kosaner J, Sonuguler S, Olgun L, Amann E. Young cochlear implant users' auditory development as measured and monitored by the LittlEARS® Auditory Questionnaire: a Turkish experience. Int J Pediatr Otorhinolaryngol, 2013; 77(8): 1359–63. https://doi.org/10.1016/j.ijporl.2013.05.036

67. May-Mederake B, Kuehn H, Vogel A, Keilmann A, Bohnert A, Mueller S, et al. Evaluation of auditory development in infants and toddlers who received cochlear implants under the age of 24 months with the LittlEARS® Auditory Questionnaire. Int J Pediatr Otorhinolaryngol, 2010; 74(10): 1149–55. https://doi.org/10.1016/j.ijporl.2010.07.003

68. Wang X, Lin Z, Guo Y, Liu Y, Zhou X, Bai J, Liu H. Correlation between cortical auditory evoked potential and auditory speech performance in children with cochlear implants. Int J Pediatr Otorhinolaryngol, 2023; 172. https://doi.org/10.1016/j.ijporl.2023.111687

69. Liu H, Jin X, Li J, Liu L, Zhou Y, Zhang J, et al. Early auditory preverbal skills development in Mandarin speaking children with cochlear implants. Int J Pediatr Otorhinolaryngol, 2015; 79(1): 71–5. https://doi.org/10.1016/j.ijporl.2014.11.010

70. Honigman T, Cushing SL, Papsin BC, Waltzman S, Woodard J, Neumann S, et al. Safety and early outcomes of cochlear implantation of Nucleus devices in infants: a multi-centre study. Trends Hear, 2024; 28. https://doi.org/10.1177/23312165241261480

71. Yidi L, Yue L, Xinyu W, Jiaying H, Xin N, Haihong L. Establishment of reference values for early auditory preverbal skills of children with cochlear implants. Trends Hear, 2022; 26. https://doi.org/10.1177/23312165221128435

72. Zhao Y, Li Y, Long Y, Jin X, Zheng Z, Liu Y, et al. Comparison of the development of early auditory and preverbal skills in Mandarin-speaking children with cochlear implants with and without additional disabilities. Acta Otolaryngol, 2019; 139(12): 1098–1103. https://doi.org/10.1080/00016489.2019.1670358

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