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Editor-in-Chief
Prof. Henryk Skarzynski, M.D., Ph.D., Dr. h.c. multi

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Journal of Hearing Science®

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Dear Colleagues,

As we close out another transformative year in hearing research, we are delighted to present an issue that embodies innovation, clinical insight, and interdisciplinary collaboration. Our opening hypothesis paper, "Towards a unified method of assessing early auditory development after cochlear implantation: Measuring relative delay," emphasizes the critical need for early auditory stimulation – ideally before 12 months – in children with profound sensorineural hearing loss. By reviewing various approaches to the LittlEARS auditory questionnaire and identifying challenges related to variability and intervention age, the paper introduces a novel "relative delay" metric designed to facilitate meaningful comparisons across studies and strengthen meta-analytical approaches in evidence-based practice.

This issue also features a series of original research papers that deepen our understanding of auditory function in diverse populations. One study explores extended high-frequency thresholds, auditory figure–ground discrimination, and working memory in females with polycystic ovary syndrome. Other research examines the impact of fetal hemoglobin levels on auditory discrimination in individuals with sickle cell anemia, investigates whether impeded biomechanics can influence hearing preservation in cochlear implant recipients, and evaluates the efficacy of a smartphone-based screening application for the early identification of hearing loss in school-children.

Rounding out our publication are insightful reports from two major international conferences: the 36th World Congress of Audiology in Paris and the 7th Congress of the Confederation of European ORL-HNS in Dublin.

With kind regards and greetings,

Prof. Henryk Skarzynski, M.D., Ph.D., Dr. h.c. multi

Hypothesis papers

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

Contributions:
A Study design/planning
B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

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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

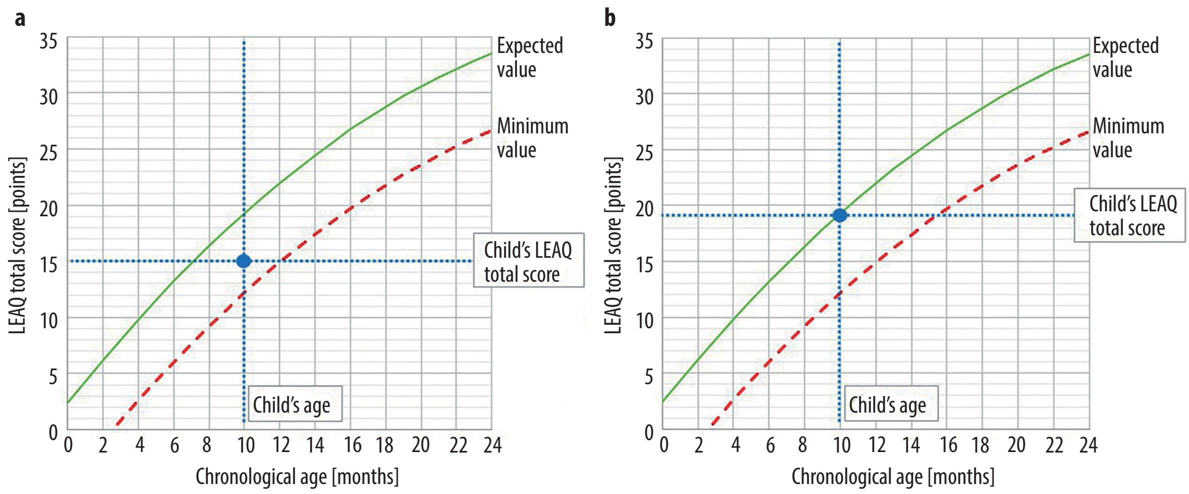

Figure 1. Interpretations of LEAQ total scores. **a**) Total score (blue dot) above minimum value (red dashed line), indicating satisfactory auditory development for a 10-month-old with a CI. **b**) Total score of 10-month-old equal to the average score of a normal hearing 10-month-old child (blue dot 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

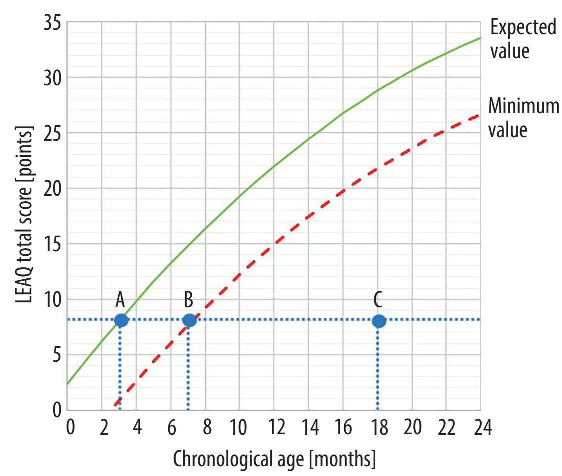

Figure 2. By itself, an LEAQ score is meaningless without knowing the child's age. For example, an LEAQ score of 8 points could come from a normal baby 3 months old (A), from a 7-month-old baby at minimum value (B), or from an 18-month-old far below the minimum (C)

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.

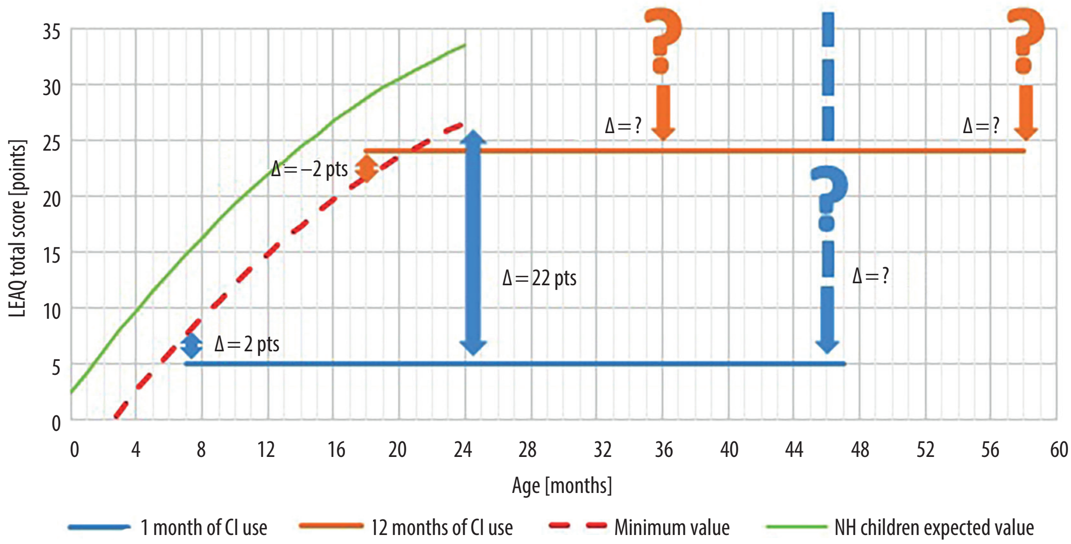

Figure 3. Relation between normative curve and average LEAQ total scores in a group of children implanted at ages ranging from 6 to 46 months. Blue line: after 1 month of CI use; orange line: after 12 months of CI use

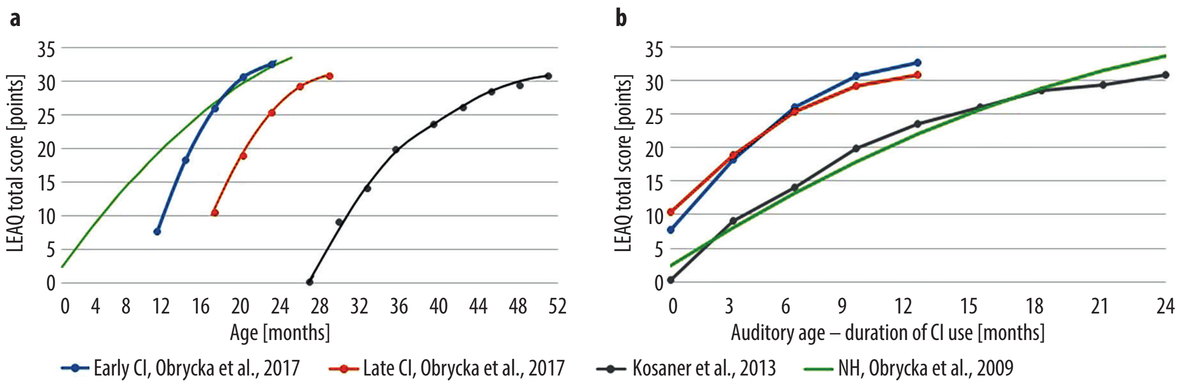

Figure 4. Two ways of plotting LEAQ total scores for groups of children implanted "early" (before 12 months of age, blue lines), "late" (12–24 months, orange lines), and "out-of-LEAQ-range" (black lines for data from Kosaner et al. 2013 [66]). **a**) Data plotted as a function of chronological age; **b**) 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

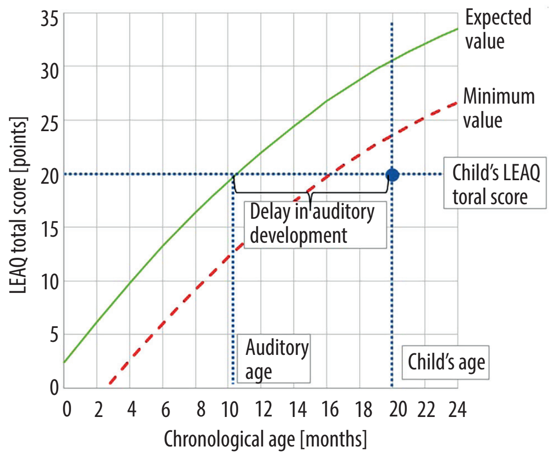

Figure 5. Interpreting LEAQ scores in terms of delay in auditory development. The example is a 20-month-old child with an LEAQ total score of 20 pts (blue dot). This same score would be achieved by a normal child at 10.5 months, meaning that in this case the auditory age is 10.5 months and the delay 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,

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

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

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

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

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:

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

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:

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

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

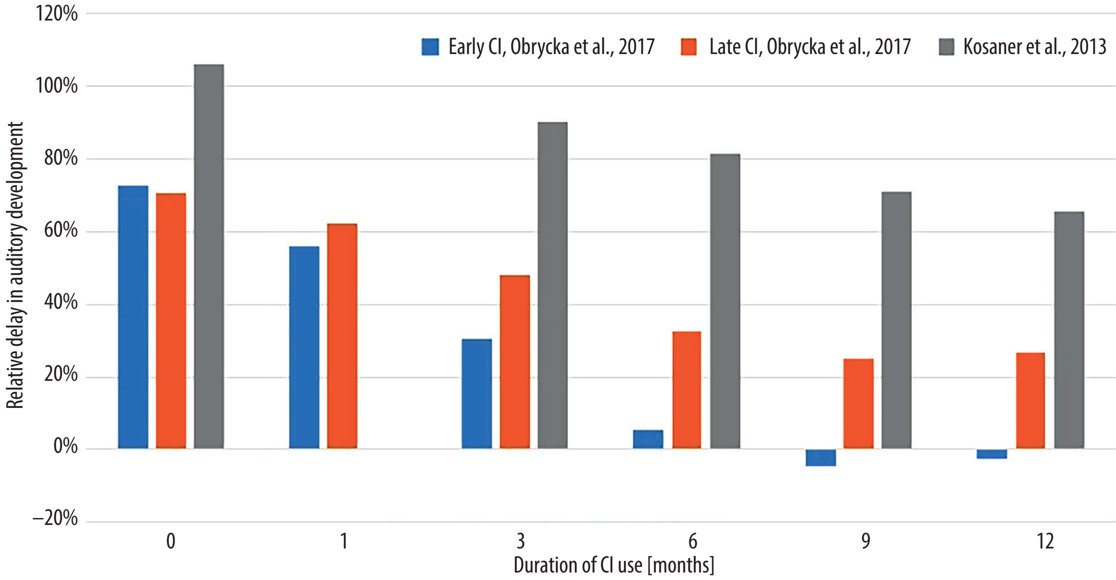

Figure 6. Relative delay in auditory development recalculated from studies by Obrycka et al. [30] and Kosaner et al. 2013 [66]

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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CROSS-SECTIONAL STUDY OF EXTENDED HIGH-FREQUENCY THRESHOLDS, AUDITORY FIGURE-GROUND DISCRIMINATION, AND WORKING MEMORY IN FEMALES WITH POLYCYSTIC OVARY SYNDROME (PCOS)

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Contributions:
A Study design/planning
B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

Abstract

Introduction: Polycystic ovary syndrome (PCOS) affects up to 10% of reproductive-age women, yet its impact on auditory function remains underexplored. This study aimed to compare auditory and cognitive functions between PCOS patients and age-matched controls.

Material and methods: Participants were 60 normal-hearing individuals aged 20–25 years, evenly split into two groups: Group 1 (control) consisted of unmarried females with regular menstrual cycles, and Group 2 (clinical group) comprised unmarried females diagnosed with PCOS. Auditory assessment involved extended high-frequency audiometry and speech perception in noise (SPIN) tests. Auditory working memory was evaluated through digit span and digit sequencing tasks.

Results: Results showed significantly poorer extended high-frequency audiometry thresholds and SPIN scores in the PCOS group compared to controls. Additionally, PCOS participants performed significantly worse on the digit span task, indicating poorer auditory working memory.

Conclusions: Extended high-frequency audiometry and reduced auditory figure-ground discrimination at low signal-to-noise ratios could potentially serve as early indicators of cochlear abnormalities at the basal end of the cochlea. Future research is needed to investigate the interplay of hormonal milieu and central processing in PCOS.

Keywords: PCOS • working memory • speech perception in noise • extended high-frequency thresholds

PRZEKROJOWE BADANIE PROGÓW W ZAKRESIE ROZSZERZONYCH WYSOKICH CZĘSTOTLIWOŚCI, DYSKRYMINACJI SŁUCHOWEJ FIGURA–TŁO I PAMIĘCI ROBOCZEJ U KOBIET Z ZESPOŁEM POLICYSTYCZNYCH JAJNIKÓW (PCOS)

Streszczenie

Wprowadzenie: Zespół policystycznych jajników (PCOS) dotyka do 10% kobiet w wieku rozrodczym, jednak jego wpływ na funkcje słuchowe jest jeszcze niedostatecznie zbadany. Niniejsze badanie miało na celu porównanie funkcji słuchowych i poznawczych między pacjentkami z PCOS a dobraną wiekowo grupą kontrolną.

Materiał i metody: Uczestniczkami badania było 60 prawidłowo słyszących kobiet w wieku 20–25 lat podzielonych na dwie równe grupy: grupa 1 (kontrolna) składała się z niezamężnych kobiet z regularnymi cyklami menstruacyjnymi, a grupa 2 (kliniczna) składała się z niezamężnych kobiet, u których zdiagnozowano PCOS. Ocena słuchowa obejmowała audiometrię w rozszerzonym paśmie wysokich częstotliwości i testy percepcji mowy w hałasie (SPIN). Słuchowa pamięć robocza została oceniona za pomocą testów rozpiętości i sekwencjonowania cyfr.

Wyniki: Wyniki wykazały istotnie niższe progi audiometrii w rozszerzonym paśmie wysokich częstotliwości i wyniki SPIN w grupie PCOS w porównaniu z grupą kontrolną. Dodatkowo osoby z PCOS osiągały istotnie gorsze wyniki w teście sekwencjonowania cyfr, co wskazuje na gorszą słuchową pamięć roboczą.

Wnioski: Audiometria w rozszerzonym paśmie wysokich częstotliwości i zmniejszona dyskryminacja słuchowa figura-tło przy niskim stosunku sygnału do szumu mogą potencjalnie służyć jako wczesne wskaźniki nieprawidłowości funkcjonowania ślimaka w obszarze jego części podstawnej. W przyszłości należałoby zbadać wzajemne oddziaływania środowiska hormonalnego i centralnego przetwarzania słuchowego w PCOS.

Słowa kluczowe: PCOS • pamięć robocza • percepcja mowy w hałasie • progi w zakresie rozszerzonych wysokich częstotliwości

Key for abbreviations	
AWM	auditory working memory
CRP	c-reactive protein
CVD	cardiovascular diseases
DPOAEs	distortion product otoacoustic emissions
EHF	extended high-frequency
IMT	intima-media thickness
LEAP-Q	Language Experience and Proficiency Questionnaire
MCRs	message to competition ratios
MOC	medial olivocochlear (reflex)
OAE	otoacoustic emissions
PCOS	polycystic ovary syndrome
QuickSIN	Quick Speech Perception in Noise (test)
SD	standard deviation
SNR	signal to noise ratio
SPIN	Speech Perception in Noise (test)
VWM	Visual Working Memory (test)
WM	working memory
WMC	working memory capacity
WRS	word recognition score

Introduction

Polycystic ovary syndrome (PCOS) is a common metabolic disorder that impacts around 10% of women in their reproductive years [1,2]. It is a chronic condition marked by wide array of clinical manifestations that can vary in severity throughout a woman's life [3,4]. Key features include irregular menstrual cycles (oligo-amenorrhea), hyperandrogenism, and polycystic ovaries [5,6]. The development and exacerbation of PCOS involve both intrinsic factors such as insulin resistance, inflammation, and altered hormone production, and extrinsic factors including environmental pollutants, epigenetic influences, diet, and stress [7,8]. Environmental and epigenetic factors play critical roles in regulating genetic expression associated with PCOS [9].

PCOS sufferers are often prone to multiple morbidities, such as coronary heart disease, type 2 diabetes, hyperinsulinemia and dyslipidemia [10–12], infertility, gestational hypertension, miscarriage or premature delivery, non-alcoholic steatohepatitis, metabolic syndrome, sleep disorders, depression, anxiety, eating disorders, and endometrial cancer [13]. Predisposing women to endothelial damage are hyperinsulinemia, insulin resistance, dyslipidemia, and low-grade chronic inflammation [11]. Diseases that cause endothelial damage often lead to early high-frequency hearing loss. Oghan and Coksuer [4] were the first to identify high-frequency hearing loss, specifically in the 4–8 kHz range, in patients with PCOS. Later, Kucur et al. [14] found that hearing impairment in PCOS patients extend to even higher frequencies (8–14 kHz).

Moreover, young PCOS cohorts often have increased carotid intima-media thickness (IMT) in comparison to those without hyperandrogenism [4]. Carotid IMT is a subclinical indicator used to evaluate atherosclerosis and cardiovascular diseases (CVD). Research has demonstrated that endocrinal and biochemical changes associated with PCOS can affect blood flow, potentially contributing to sensorineural hearing loss caused by vascular abnormalities [4]. Sundararaj et al. [15] reported that endocrinal and biochemical changes, hyperandrogenism, cardiovascular problems, insulin resistance, and endothelial damage impact auditory function in females with PCOS.

Studies indicate that individuals with PCOS are more likely to experience hidden hearing loss at high frequencies (8–20 kHz) than at lower frequencies (0.25–4 kHz) [14]. This type of hearing loss, due to cochlear damage, can hinder speech understanding in noisy environments. Shaw et al. [16] suggested that reduced speech perception in the presence of noise may result from extended high-frequency (EHF) hearing loss. Motlagh et al. [17] found that young adults in their 20s with normal standard audiometric results still showed reduced speech perception in challenging conditions due to EHF hearing loss. Thus, it is essential to study how high-frequency hearing loss affects auditory figure-ground discrimination (speech perception in noise, SPIN) in young women with PCOS. Fluctuations in ovarian hormones like estrogen and progesterone can affect inner ear homeostasis and overall auditory function. Estrogen, in particular, is known to protect the auditory system by activating the medial olivocochlear (MOC) reflex [18,19]. Kumar et al. [19] compared MOC function between PCOS patients and age-matched males using QuickSIN (quick speech perception in noise test) in Malayalam [20], finding that PCOS participants had significantly poorer scores.

High-frequency hearing loss is well-documented in females with PCOS [4,14,15]. Further research is needed to investigate auditory figure-ground discrimination ability in young women with PCOS. A study by Apeksha et al. [21] explored this in middle-aged women with PCOS, highlighting the need for additional research focused on younger populations to better understand the relationship between EHF hearing loss and auditory figure-ground discrimination.

In the general population, reproductive and metabolic disturbances each affect cognitive function separately [22]. Physiological and pathological alterations in the levels of testosterone and estrogen hormones can lead to changes in cognitive function [23,24]. In women with PCOS, cognitive function may be influenced by hyperandrogenism and hyperestrogenism [12]. However, there are limited studies that have explored cognitive function in women with PCOS [22,25–28]. Barnard et al. [25] conducted an online study involving 221 women with PCOS (some of whom were on antiandrogenic treatment) and 442 control participants, and found that women with PCOS had notably slower reaction times and reduced word recognition. Additionally, those undergoing antiandrogenic treatment showed improved performance compared to those not receiving such treatment. Schattmann and Sherwin [22] found that women with high free testosterone levels from PCOS performed less effectively on tasks typically associated with female-oriented skills, such as verbal memory, fluency, and visuospatial working memory compared to control women. However, these women did not exhibit superior performance on tasks generally favored by men.

Working memory (WM) plays a significant role in higher cognitive functions [29–32]. The hippocampus, situated in the medial temporal lobe, is rich in androgen and estrogen receptors and plays a vital role in WM [33]. Working memory capacity (WMC) can be evaluated through simple tasks (forward, backward, ascending, and descending digit span, along with visual and spatial span) as well as complex tasks (including reading span, operational span, rhyme judgment, and visual letter monitoring) [34]. Sundararaj et al. [15] assessed auditory working memory in 20 women with polycystic ovarian syndrome using digit span and sequencing tasks. The study found that women with PCOS performed poorly on backward digit span and both ascending and descending digit sequencing tasks. These deficits may be linked to hormonal irregularities such as hyperandrogenism and imbalances in testosterone and estrogen levels in PCOS [35,35a].

WM encompasses the selection and processing of stimuli from a single sense as well as the integration of information from different senses, such as vision and hearing [36,37]. Research combining these sensory modalities is of considerable scientific interest. To evaluate higher cognitive functions, the brain might have to adjust and integrate information from both visual and auditory inputs [38]. We hypothesized that impaired auditory working memory due to hormonal variation might also be reflected in the visual modality. Therefore, this study examined the auditory and visual working memory of women with PCOS and compared them to age-matched controls. The objectives of the study were to assess EHF thresholds, speech perception in noise (SPIN), as well as auditory and visual working memory in individuals with PCOS.

Material and methods

Using the purposive sample method we recruited 60 final-year graduate students aged between 20–25 years from our allied health science university. These participants had normal/corrected vision, hearing, and intelligence and had no history of mental or neurological diseases. All participants provided written consent and voluntarily agreed to take part.

Inclusion criteria

All the participants were bilingual individuals with Malayalam as the primary language and English as a second language. Language proficiency was assessed using the Language Experience and Proficiency Questionnaire [39] (LEAP-Q score > 80% for Malayalam and English). Participants were evenly divided into two groups, with 30 in each. Group 1, the control group, comprised unmarried females with regular menstrual cycles with a mean age of 22.3 years ($SD = 1.57$), while Group 2, the clinical group, included unmarried females diagnosed with PCOS with a mean age of 22.0 years ($SD = 1.56$). All participants had air conduction thresholds < 15 dB HL and bone conduction thresholds < 10 dB HL at octave frequencies, as well as speech recognition scores of > 90% in quiet. Bilateral ipsi and contra reflexes were within normal limits, and participants also had type A tympanograms using a 226 Hz probe tone. DPOAEs were detected in all participants, with signal to noise ratio (SNR) > 6 dB over 2–5 kHz.

Participants in the clinical group were recruited after being diagnosed with PCOS according to Rotterdam's criteria [40]. All selected individuals had been diagnosed with PCOS for at least 3 years prior to the study and were currently receiving treatment. Despite their medication, all participants exhibited clinical symptoms of PCOS such as irregular menstrual cycles, hirsutism, and acne during the data collection period.

Exclusion criteria

History or complaints of loss of hearing in one or both ears, neurological diseases, endocrine diseases such as diabetes, androgen-secreting tumors and thyroid dysfunctions, hypertension, exposure to ototoxic drugs, noise exposure, autoimmune diseases, and intake of any medications which could alter sex hormones.

Ethical standards

Participants were briefed on the study's objectives and procedures before commencement, and informed consent was obtained from each participant. All procedures were non-intrusive and adhered to the Ethics committee of the Institute (approval number AWH/EC/02/2022) and complied with the Declaration of Helsinki.

Instrumentation

A calibrated Maico MA 42 dual-channel diagnostic audiometer in conjunction with TDH 39 headphones fitted with MX-41/AR cushions and a Radioear B-71 bone vibrator were used to perform pure tone and speech audiometry. A Malayalam high-frequency wordlist was delivered to the ear with better hearing from the same audiometer during an auditory figure-ground discrimination test. The middle ear condition was evaluated using a GSI Tympstar Pro middle ear analyzer. A Smart DPOAE Intelligent Hearing System was used to measure DPOAEs. Auditory and visual working memory was evaluated using Smriti Shravan software, a customized tool for assessing working memory [40a]. The stimulus was delivered via a Dell Inspiron Core i3 laptop calibrated with TDH-49 headphones.

Test environment

Tests were conducted in an acoustically treated and well-lit air-conditioned room with ambient noise levels within ANSI S.3 (1991). Pure tone audiometry and SPIN tests at various MCRs were performed in a two-room setup, while OAE measurements and working memory assessments were conducted in a single room.

Procedure

Hearing evaluation

Each subject underwent an otoscopic examination to ensure the absence of earwax, foreign objects, and abnormalities in the tympanic membrane. Pure tone and speech audiometry tests were conducted using the modified Hughson and Westlake procedure [41] to determine thresholds. Spondees were used for speech reception threshold assessment [42], while monosyllabic wordlists were employed to measure speech identification scores [42]. Stimuli were presented in real-time at 40 dB SL. DPOAEs were recorded using a Maico Ero Scan.

Acquisition of EHF thresholds

Sennheiser HAD 300 circumaural headphones were used to provide pure tones to measure EHF hearing thresholds at 9, 10, 11.2, 12.5, 14, and 16 kHz. The thresholds were obtained using the modified Hughson and Westlake technique.

SPIN test

Speech perception in noise was assessed at different message competing ratios (–5, 0, and +5 dB MCRs), and a Malayalam high-frequency word list [43] was utilized alongside multitalker babble controlled by custom-written Matlab code. The stimuli were delivered to the right ear at a comfortable level from a Dell Inspiron Core i3 laptop through a clinical audiometer. The right ear advantage is not specific to the dichotic condition and has been observed with monaural presentations in normal healthy adults [44]. Among healthy normal children and adults, the word recognition score (WRS) in the right ear has been reported to be slightly better than that of the left ear in the noise condition [45]. Hence the auditory figure-ground discrimination test was only administered to the right ear.

Recordings were conducted in a sound-proof room, adhering to noise level guidelines specified in ANSI S3.1-1991. Each word list, randomized for presentation, was spoken by a female native speaker of Malayalam (who was a voice professional) and recorded in Praat software. The speaker was instructed to pronounce words naturally, clearly, and with neutral intonation while maintaining consistent vocal effort. Post-recording, each word was normalized to 0 dB using Adobe Audition v. 3.0. Additionally, a 1 kHz calibration tone normalized to 0 dB was generated in Adobe Audition and added at the beginning of each word list.

Working memory tests

The assessment of auditory and visual working memory utilized Smriti Shravan software, a tool designed for this purpose [40a].

Auditory working memory

Stimuli consisted of sets of digits from the auditory module of the software, which were presented simultaneously to both ears of each participant. In the forward span task, participants were asked to arrange the digits in the exact order they were heard in English. Similarly, in the backward, ascending, and descending spans, participants were instructed to sequence the digits in reverse, ascending, and descending orders, respectively. During each trial, digits were presented sequentially with a 1 s interval between each digit. After completing a full trial, the participant had 5 s to input the heard digits in a particular sequence according to each task's instructions. Before the actual testing, each task included a trial for familiarization, with instructions displayed on the computer screen. The examiner provided supplementary verbal instructions as required.

The number of digits in each trial depended on the participant's responses. Correct responses resulted in an increase of one digit in subsequent trials, while incorrect responses led to a reduction of one digit. The stimulus trials included digits from 1 to 9, with the exception of 7, which had a longer duration than the others. Stimuli were presented through a Dell Inspiron Core i3 laptop paired with Sennheiser HD 206 headphones, at an intensity of 40 dB SL (relative to the speech recognition threshold) for all participants, in a quiet, distraction-free environment. Scoring utilized a one-up, one-down adaptive procedure, and the final score was determined as the average of the midpoints of the last four reversals. The software computed scores for each task and the scores from the midpoint of the last three reversals were extracted for ascending, descending, forward, and backward spans. These scores were then compared between the groups using suitable statistical methods. The software computed the scores achieved for each task. Scores from the midpoint of the last three reversals were obtained for ascending, descending, forward, and backward spans, and were compared between groups using appropriate statistical methods.

Visual working memory

In this task, participants were required to memorize and subsequently recall visual stimuli (numbers) presented in the forward, backward, ascending, and descending span tasks using the visual module. The remaining test procedure and scoring were identical to those described earlier. Test results were automatically recorded and saved in an Excel spreadsheet.

The obtained data were tabulated and analyzed by using Statistical Package for Social Sciences (SPSS, v. 26.0). A Shapiro–Wilk test of normality was administered to verify the normal distribution of the obtained data and findings showed that data was not normally distributed. Hence non-parametric tests were administered. Descriptive statistics were done to estimate the mean and standard deviation (SD) of extended high-frequency audiometry, SPIN, and working memory assessment for all participants. Mann–Whitney *U*-tests were done to compare the EHF thresholds for right and left ears between groups. Mann–Whitney *U*-tests were conducted to test whether there was any significant difference between the experimental and control groups for SPIN scores at various MCRs. The auditory working memory results for digit span and digit sequencing were compared using Mann–Whitney *U*-tests between control and experimental groups.

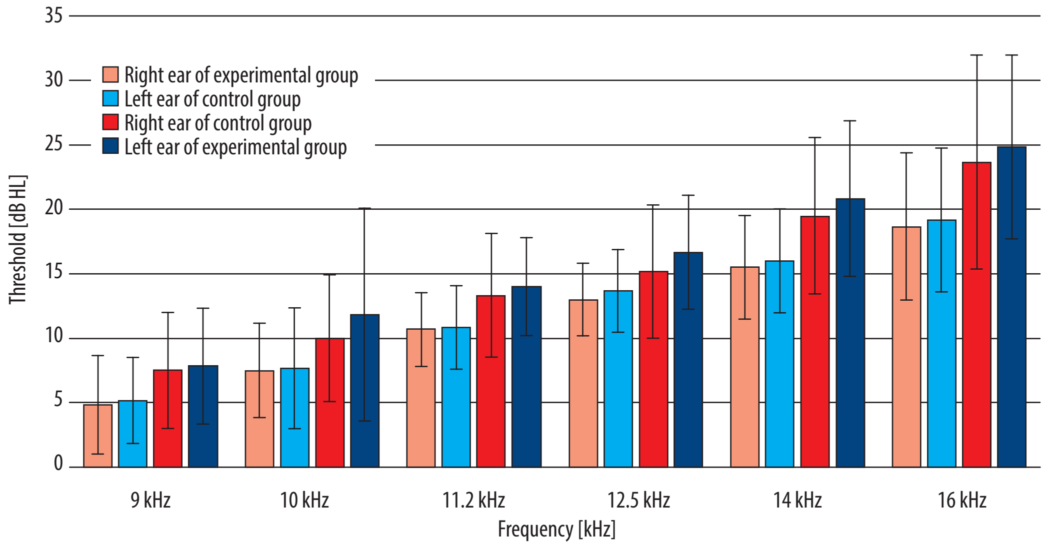

Figure 1. Mean and standard deviation of EHF thresholds in control and experimental groups

Table 1. Comparison of EHF thresholds across the groups

	9 kHz	10 kHz	11.2 kHz	12.5 kHz	14 kHz	16 kHz
Mann–Whitney *U*	299.5	311	285.5	345.5	272.5	290
Z	–2.4	–2.2	–2.6	–1.7	–2.8	–2.4
Asymp. Sig. (2-tailed)	**.019**	**.030**	**.009**	.090	**.005**	**.014**

Results

EHF thresholds

Figure 1 shows the mean and standard deviation of EHF thresholds in PCOS females (saturated colours) compared to age-matched healthy controls (light colours). PCOS females returned significantly poorer mean scores in both ears than in controls, although both groups showed a right ear advantage.

Mann–Whitney *U*-tests were used to compare the EHF thresholds between groups across frequencies. A statistically significant difference was obtained ($p < 0.05$) across all frequencies except at 12.5 kHz (**Table 1**).

Speech Perception in Noise Test (SPIN)

Figure 2 shows the mean and standard deviation of SPIN scores across different MCR levels (5, 0, –5 dB SNR) in PCOS females compared to age-matched healthy individuals. Across all MCRs, PCOS females exhibited lower mean scores, with statistically significant differences observed, particularly at 0 and –5 dB MCR.

Mann–Whitney *U*-tests were administered to check if there were any significant differences between scores of SPIN at 5, 0, and –5 dB MCR in both the control group and the experimental group. A statistically significant difference

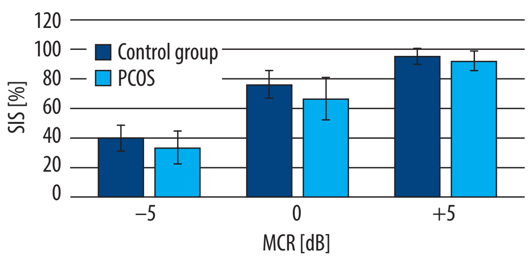

Figure 2. Means and standard deviations of speech in noise (SPIN) in both groups

was obtained in all MCRs except at +5 dB for both the control group as well as the experimental group (**Table 2**).

Working memory assessment

Auditory digit span and sequencing tests

Figure 3 depicts the mean and *SD* of forward digit span tests, backward digit span, ascending sequence, and descending sequence test of both groups. Females with PCOS had lower mean scores overall, with the forward digit span test yielding the highest mean score and the descending sequence test the lowest. This was true for both groups.

Table 2. Comparison of different MCRs in both groups

	SPIN scores at various MCRs [%]		
	–5 dB	**0 dB**	**+5 dB**
Mann–Whitney *U*	298	280	333
Z	–2.3	–2.5	–1.8
Asymp. Sig. (2-tailed)	**0.023**	**0.011**	0.071

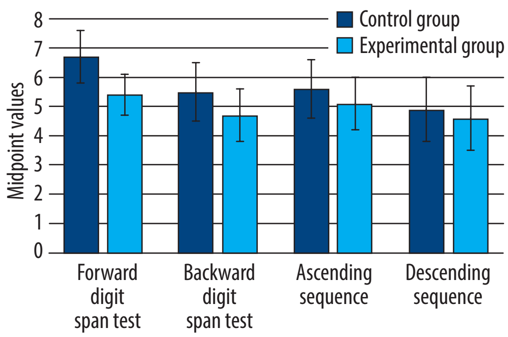

Figure 3. Mean and standard deviation of auditory working memory tests in both groups

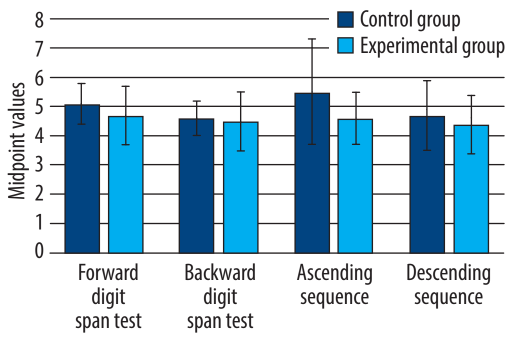

Figure 4. Mean and standard deviation of visual working memory tests in both groups

Table 3. Comparison of auditory working memory tests between both groups

	Forward digit span test	Backward digit span test	Ascending sequence	Descending sequence
Mann–Whitney *U*	119	239.5	335.5	360.5
Z	–4.9	–3.1	–1.7	–1.3
Asymp. Sig. (2-tailed)	**< 0.001**	**0.002**	0.090	0.186

Table 4. Comparison of visual working memory tests between both groups

	Forward digit span test	Backward digit span test	Ascending sequence	Descending sequence
Mann–Whitney *U*	319.5	437.5	294.0	412.0
Z	–1.930	–0.185	–2.307	–0.562
Asymp. Sig. (2-tailed)	0.054	0.853	**0.021**	0.574

Comparison of AWM between groups

Mann–Whitney *U*-tests were administered, and a statistically significant difference was present only for forward and backward digit span tests (**Table 3**).

Visual digit span and sequencing tests

Visual working memory was evaluated using digit span and sequencing tasks, analogous to those used for auditory assessment. Females with PCOS had overall lower scores, with the forward digit span test producing the highest scores and the descending sequence test the lowest. In the control group, the ascending sequence test had the highest scores, while the backward digit span test had the lowest. Mean scores are plotted in **Figure 4**.

Comparison of VWM tests between groups

Mann–Whitney *U*-tests were administered and only the ascending sequence span tests showed a statistically significant difference (**Table 4**).

Correlation between AWM and VWM in PCOS females

To evaluate the correlation between auditory working memory with visual working memory in the experimental

Table 5. Correlation between auditory and visual working memory in the experimental group

			AWM				VWM			
			F.S	**B.S.**	**A.S.**	**D.S.**	**F.S**	**B.S.**	**A.S.**	**D.S.**
AWM	F.S.	correlation coefficient	1.00	0.47	–0.16	–0.36	–0.10	0.17	–0.12	–0.29
		Sig. (2-tailed)		**0.01**	0.39	0.05	0.60	0.36	0.53	0.11
	B.S.	correlation coefficient	0.47	1.00	–0.07	0.01	–0.19	0.09	–0.02	–0.36
		Sig. (2-tailed)	**0.01**		0.71	0.94	0.32	0.63	0.90	**0.048**
	A.S.	correlation coefficient	–0.16	–0.07	1.00	0.60	–0.26	–0.13	0.48	0.49
		Sig. (2-tailed)	0.39	0.71		**< 0.001**	0.17	0.49	**0.01**	**0.01**
	D.S.	correlation coefficient	–0.36	0.01	0.60	1.00	–0.23	–0.17	0.49	0.53
		Sig. (2-tailed)	0.05	0.94	**< 0.001**		0.23	0.36	**0.01**	**< 0.001**
VWM	F.S.	correlation coefficient	–0.10	–0.19	–0.26	–0.23	1.00	0.68	–0.13	–0.09
		Sig. (2-tailed)	0.60	0.32	0.17	0.23		< 0.001	0.51	0.62
	B.S.	correlation coefficient	0.17	0.09	–0.13	–0.17	0.68	1.00	0.11	–0.11
		Sig. (2-tailed)	0.36	0.63	0.49	0.36	**< 0.001**		0.55	0.56
	A.S.	correlation coefficient	–0.12	–0.02	0.48	0.49	–0.13	0.11	1.00	0.50
		Sig. (2-tailed)	0.53	0.90	**0.01**	**0.01**	0.51	0.55		**0.01**
	D.S.	correlation coefficient	–0.29	–0.36	0.49	0.53	–0.09	–0.11	0.50	1.00
		Sig. (2-tailed)	0.11	**0.048**	**0.01**	**< 0.001**	0.62	0.56	**0.01**	

Abreviations: AWM, auditory working memory; VWM, visual working memory; F.S., forward span; B.S., backward span; A.S., ascending span; D.S., descending span

group, Spearman's rank correlation was used. **Table 5** shows the correlation between auditory and visual working memory and it shows that there is a low positive significant correlation between forward and backward digit span tests in auditory working memory (correlation = 0.47, $p = 0.01$), ascending and descending sequence span in auditory working memory (correlation = 0.60, $p < 0.001$), ascending sequence span in both auditory and visual working memory (correlation = 0.48, $p = 0.01$), ascending sequence span in auditory and descending sequence span in visual working memory (correlation = 0.49, $p = 0.01$), descending sequence span in auditory and ascending sequence span in visual working memory (correlation = 0.49, $p = 0.01$), descending sequence span in both auditory and visual working memory (correlation = 0.53, $p = 0.01$), forward and backward digit span in both visual working memory (correlation = 0.68, $p = 0.01$). There is a low negative significant correlation between backward digit span tests in auditory and descending sequence span in visual working memory (correlation = –0.36, $p = 0.048$).

Discussion

The study aimed to assess hearing and cognition in PCOS individuals. EHF audiometry and SPIN tests were used to assess hearing, while cognition was evaluated using tests for auditory and visual working memory.

EHF audiometry

Results of the current study revealed that young women with PCOS had poorer EHF thresholds (9–16 kHz), which is in accordance with previous findings. Both Oghan and Coksuer [4] and Kucur et al. [14] reported that the hearing thresholds of their PCOS groups were higher at EHF compared to controls. The cause of the loss could be insulin resistance, hyperandrogenemia, or elevated serum CRP as an inflammatory marker of PCOS.

A recent study on PCOS cohorts aged 18–40 years reported similar findings [21]. Elevated thresholds were attributed to vascular obstructions in the arteries which feed the inner ear with oxygen. Hearing in the low and mid frequencies may improve if blood flow returns to normal, but hearing in the higher frequencies may not [46]. Increased carotid intima-media thickness in young women with PCOS results in endocrinal and biochemical changes, which could affect the blood flow and potentially contribute to hearing loss. The current study on young PCOS females observed signs of hyperandrogenism such as hirsutism, acne, alopecia, and irregular menstrual cycles, and there was elevated body weight even under medication. Turan et al. [47] proposed that a rise in testosterone levels, primarily associated with hirsutism, could contribute to elevated hearing thresholds.

SPIN

Poorer SPIN scores for the Malayalam high-frequency word list were obtained at lower SNRs (−5 and 0 dB) by the young PCOS females. Apeksha et al. [21] noted reduced SPIN scores among PCOS-afflicted women at −3, −6, and −9 dB SNR. The hidden high-frequency hearing loss resulting from cochlear damage might therefore hinder the perception of high-frequency words in the presence of noise. Other researchers have suggested that fluctuations in estrogen levels in the ovaries may influence SPIN scores, while progesterone may also have an impact [48]. Thus, the reduced SPIN scores in PCOS females may stem from fluctuations in female reproductive hormones. This study supports the suggestion that the diminished SPIN performance in PCOS cohorts could result from disturbances in hormonal equilibrium, together with possible auditory and neural impairments linked to vascular constriction.

AWM in PCOS females

Women with PCOS demonstrated lower mean scores on auditory working memory tests such as digit span and sequence tasks. A statistically significant difference was noted in the forward and backward digit span test whereas no such difference was observed in the sequence tasks. A previous study [15] reported a significant difference in all tasks except the forward task. From these findings, it is clear that auditory working memory is affected in these individuals, and can possibly be attributed to elevated levels of testosterone and estrogen associated with hyperandrogenism in PCOS. An alternative explanation suggests that women with PCOS show increased activation in brain regions such as the superior and inferior parietal lobes and the superior temporal lobe. However, such an increase in activation is not associated with working memory storage or attentional processes compared to women in the control group [49].

The lack of a significant difference in the sequencing task between groups might be attributed to their younger age and ongoing hormonal treatment. Collectively, these factors help mitigate significant declines in working memory in PCOS cohorts, since Soleman [49] noted that hormonal treatment could potentially alleviate memory issues and concluded that antiandrogenic treatment improves cognitive performance.

VWM in PCOS females

The mean and standard deviation of visual working memory digit span and sequence tasks revealed that females had poorer mean scores, even though the difference was statistically significant only in the ascending sequence test. Lai et al. [50] conducted a cross-sectional study on 21 PCOS cohorts and reported decreased activities of brain regions responsible for visuospatial working memory such as the left inferior temporal and occipital gyrus. The literature suggests that the parietal region acts as a storage buffer for visual information [51]. Reduced efficiency in working memory processing may lead to the need for compensatory parietal activity. But this may not be observed in women with PCOS, due to suboptimal processing. The above literature supports the contention that VWM is affected in young PCOS females, even though in the current study there was no significant differences across various digit tasks. The lack of a significant difference may be attributed to the inherent difficulty and time needed to complete each task, which led to generally low scores across both the experimental and control groups. Future research may aim to reduce these difficulties. Despite these limitations, this study serves as an initial exploration into understanding the impact of PCOS on visual working memory.

The correlation observed between auditory and visual working memory tasks suggests that the cognitive effects of PCOS are not isolated to specific modalities but rather involve broader cognitive domains. Future research could delve deeper into the intricate interplay between hormonal balance and central cognitive processing in this complex condition.

There are a number of limitations in the current study. The result of visual working memory tasks can be taken as preliminary findings from which a future study could be designed with greater control on extrinsic variables. Also, this study cannot say whether specific hormonal treatments might mitigate cognitive deficits.

Conclusions

Young women diagnosed with PCOS exhibited poorer extended high-frequency thresholds in both ears. Furthermore, their speech discrimination scores in noisy environments were also adversely affected. Cognitive function was assessed with tasks that evaluated working memory in both the auditory and visual domains, and revealed lower performance in PCOS. Moreover, extended high-frequency audiometry and reduced auditory figure-ground discrimination at low signal-to-noise ratios could potentially serve as early indicators of cochlear abnormalities at the basal end of the cochlea. Future research is needed to investigate the interplay of hormones and central processing in PCOS.

Statements and declarations

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EFFECT OF FETAL HEMOGLOBIN LEVEL ON AUDITORY DISCRIMINATION IN INDIVIDUALS WITH SICKLE CELL ANEMIA

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Contributions:
A Study design/planning
B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

Abstract

Introduction: Sickle cell anemia (SCA) is an inherited disorder characterized by hemolysis and vaso-occlusive crises. Hydroxyurea (HDU) is the primary drug available for SCA which increases the concentration of fetal hemoglobin (HbF) and has shown clinically beneficial effects ranging from 15 to 20% on sickle cells. The present study aimed to investigate the impact of HbF level on auditory discrimination ability and speech perception in noise in normal hearing individuals with SCA under HDU treatment.

Material and methods: A between-subject design was used for group comparison. Non-random purposive sampling was used to select participants from a local Hindi-speaking community. A total of 66 normal-hearing adults diagnosed with SCA and under medication with HDU in the age range of 18–40 years were divided into three groups based on HbF%. Auditory discrimination for frequency (DLF), intensity (DLI), and duration (DDT) were evaluated at 500 Hz and 4000 Hz along with SPIN at 0 dB SNR using the MLP toolbox in Matlab.

Results: Comparing the three HbF groups, Kruskal–Wallis tests revealed no significant difference in auditory discrimination based on either DLF, DLI, DDT, or SPIN scores.

Conclusions: The present study indicated no impact on the auditory system of raised HbF level in SCA individuals. The findings suggest that in such individuals not every system in the body is affected equally by raised HbF levels.

Keywords: fetal haemoglobin (HbF) • difference limen for frequency • difference limen for intensity • duration discrimination test • speech perception in noise

WPŁYW POZIOMU HEMOGLOBINY PŁODOWEJ NA DYSKRYMINACJĘ SŁUCHOWĄ U OSÓB Z NIEDOKRWISTOŚCIĄ SIERPOWATOKRWINKOWĄ

Streszczenie

Wprowadzenie: Niedokrwistość sierpowatokrwinkowa (SCA) jest dziedzicznym zaburzeniem charakteryzującym się hemolizą i kryzysami naczyniowo-okluzyjnymi. Hydroksymocznik (HDU) jest podstawowym lekiem dostępnym w leczeniu SCA, który zwiększa stężenie hemoglobiny płodowej (HbF) i klinicznie wykazał korzystny wpływ na krwinki sierpowate w zakresie od 15% do 20%. Niniejsze badanie miało na celu zbadanie wpływu poziomu HbF na zdolność dyskryminacji słuchowej i percepcję mowy w hałasie u osób z normą słuchową z SCA leczonych HDU.

Materiał i metody: Do porównania grup zastosowano model międzyosobniczy. Nielosowy, celowy dobór próby został wykorzystany do wybrania uczestników z lokalnej społeczności mówiącej w języku hindi. Łącznie 66 osób dorosłych ze słuchem w normie, z diagnozą SCA i leczonych HDU, w wieku 18–40 lat podzielono na trzy grupy na podstawie HbF%. Dyskryminacja słuchowa dla częstotliwości (DLF), intensywności (DLI) i czasu trwania (DDT) została oceniona dla częstotliwości 500 Hz i 4000 Hz wraz z SPIN przy 0 dB SNR z użyciem biblioteki MLP (Matlab).

Wyniki: Porównanie trzech badanych grupy z wykorzystaniem testów Kruskala–Wallisa nie wykazało istotnej różnicy w dyskryminacji słuchowej opartej na wynikach DLF, DLI, DDT i SPIN.

Wnioski: Niniejsze badanie nie wykazało wpływu podwyższonego poziomu HbF na układ słuchowy u osób z SCA. Wyniki sugerują, że u takich osób nie każdy układ w organizmie jest w równym stopniu dotknięty podwyższonym poziomem HbF.

Słowa kluczowe: hemoglobina płodowa (HbF) • granica różnicy częstotliwości • granica różnicy natężenia • test dyskryminacji czasu trwania • percepcja mowy w hałasie

Key for abbreviations	
DDT	duration discrimination threshold
DLF	difference limen for frequency
DLI	difference limen for intensity
Hb	hemoglobin
HbC	hemoglobin C
HBB	β-hemoglobin gene
HbF	fetal hemoglobin
HbS	sickle hemoglobin molecule
HbSC	sickle cell–HbC
HbSS	sickle hemoglobin molecule polymerization
HDU	hydroxyurea
IQR	interquartile range

Key for abbreviations	
MLP	maximum likelihood procedure
PTA	pure tone audiometry
RBCs	red blood cells
SIS	speech identification score
SCA	sickle cell anemia
SCD	sickle cell disease
SNR	signal-to-noise ratio
SPIN	speech perception in noise
SPSS	Statistical Package for the Social Sciences
SRT	speech recognition threshold
VOC	vaso-occlusive crises
WHO	World Health Organization

Introduction

Sickle cell anemia (SCA) is an inherited hematological disease affecting humans. SCA is caused by mutations in the β-hemoglobin gene (*HBB*) and its epidemiology, pathophysiology, and clinical complications have been recently reviewed [1]. The two most common genotypes of this disease are homozygosity for the *HBB* Glu6Val mutation (HbS; rs334) called sickle cell anemia (HbSS), and compound heterozygosity for HbS and HbC (Glu6Lys) mutations, called sickle cell–HbC (HbSC) disease. Among all SCD, SCA is the most severe and well-known type of sickle cell disease [1]. Clinically, there is progressive multiorgan failure, including the auditory system, and in severe cases increased mortality. The highest prevalence is in West Africa, India, the Mediterranean region, and the Middle East [2].

The two main features of SCA (an inherited globin chain disorder) are hemolysis and vaso-occlusive crises (VOC). The sickle hemoglobin (HbS) molecule is prone to change into stiff, elongated polymers in a deoxygenated state due to a defect in the *HBB* gene. Initially, sickle erythrocytes go through a cyclical process in which they alternate between their typical biconcave shape and an abnormal crescent shape which they acquire under low oxygen pressure. Eventually, the shift becomes irreversible, and the sickle erythrocytes acquire a permanent sickle shape, promoting hemolysis and VOC [2].

The auditory system is supplied by the labyrinthine artery, a branch of the anterior inferior cerebellar artery. Any VOC incidence before or at this level could affect the entire blood flow in the inner ear (the organ of Corti). Due to microvascular occlusion incidents, which can compromise oxygenation through the labyrinthine artery to the cochlea [3], ischemia of the highly metabolic cochlea and organ of Corti could result in decreased oxygenation of the stria vascularis and failure to maintain the electrochemical gradient of endolymph, which is crucial for inner and outer hair cell function [4]. Vaso-occlusion can also contribute to labyrinthine hemorrhage and labyrinthitis ossificans, which appear to be more common in patients with HbSC and HbSS genotypes, respectively [5]. Also, retrocochlear changes, i.e. functional changes in neural condition of auditory nerves, have been reported in some previous studies [6,7]. VOC has also found to affect the cerebral hemispheres and lead to neurological complications such as strokes, causing possible auditory processing deficits [8]. These multiple-level changes in the auditory system might compromise auditory psychophysical processing, including frequency, intensity, and duration discrimination, as well as ordering-related tasks, which together may result in overall poorer speech understanding.

The pathophysiology of SCA causes multiorgan damage and various acute symptoms. Early therapy with oral medication is useful in reducing the kinetics of HbSS polymerization and chronic complications like pain, acute chest syndrome, acute stroke, aplastic crises, infection with fever, priapism, ocular complications, avascular necrosis, and leg ulcers. A lifelong affliction of hemolytic anemia ultimately requires blood transfusions [9]. Hydroxyurea (HDU) is the primary drug available for SCA and remains the first line of therapy. For SCA patients, changing sickle erythrocyte kinetics is the aim of therapy.

HDU is a ribonucleotide reductase inhibitor which raises fetal hemoglobin (HbF) levels. It raises the quantity of erythrocytes containing HbF as well as its intracellular concentration. Furthermore, HDU decreases leukocyte and reticulocyte counts in circulation, increases red blood cell volume, and decreases deformability. It thus enhances blood flow in capillaries, and modifies adhesion molecule expression, thus averting vaso-occlusive crises.

According to clinical observations, increased HbF concentrations help treat SCA [10]. Laboratory research indicates that fetal hemoglobin levels of at least 15% to 20% may be necessary for a therapeutic benefit [11]. However, in a study of patients not receiving treatment any increase over 4% might be advantageous [12]. The possible beneficial circulatory changes post HDU therapy might have a direct or indirect impact on auditory system function at different levels (retrocochlear or auditory cortical regions). This may provide better overall auditory processing and speech outcomes.

Need for the study

Due to circulatory alterations, the pathophysiology of SCA results in a variety of clinical manifestations [13].

Table 1. HbF [%], gender distribution, and age of the SCA participants divided into three HbF groups

Group HbF [%]	HbF [%]				Gender			Age			
	Mean	*SD*	Median	IQR	Male	Female	Total	Mean	*SD*	Median	IQR
Group 1 10–15%	12.37	1.79	13.10	3.5	16	1	17	25.12	6.17	24.11	11.00
Group 2 15–20%	16.92	1.22	16.95	1.8	15	11	26	27.42	7.82	30.50	16.00
Group 3 > 20%	22.93	2.89	21.80	3.0	10	13	23	26.26	7.47	26.00	14.00

As an end-organ system, the auditory system is especially susceptible to VOC events. An SCA crisis could result in stasis of the inner ear's supply channel, so that the organ of Corti and stria vascularis become blocked and outer hair cells die. This can lead to permanent inner ear damage, as reported in previous studies [14]. A fully functioning inner ear and auditory system is crucial for normal hearing and processing of frequency, intensity, and temporal discrimination [15].

SCA individuals under treatment with HDU show varying levels of HbF. Increased HbF levels are reported to be useful in preventing vaso-occlusive crises by reducing the kinetics of HbSS polymerization, raising the volume of RBCs (which reduces their deformability and improves blood flow through capillaries), and altering the expression of adhesion molecules [10]. Thus, studying the effect of HbF level and possible alteration of different auditory discrimination abilities in such individuals might give a better insight into microcirculatory changes in cochlear and retro-cochlear regions. This may also help in understanding target HbF levels.

Studying effects of HbF level might be a tool for forecasting how to safeguard the auditory system from adverse effects of vaso-occlusion events. It might also help in understanding the dose correction for HDU to maintain the required level of HbF% for better outcomes, since reducing vaso-oclusive crisis events lead to a better quality of life in SCA individuals.

Nevertheless, there is no one-to-one correlation between the severity of SCA and HbF percentage. Classification of SCA is therefore not based on HbF, because HbF% could be just one independent variable determining the health of an SCA individual [11]. Thus, it is of some importance to study the impact of HbF% on auditory perception, an aspect that has not yet been explored.

The present study aims to investigate the impact of HbF level on auditory discrimination ability and speech perception in noise in individuals with sickle cell anemia who have normal hearing sensitivity.

Material and methods

A non-experimental standard group comparison method [16] was employed to achieve the aim of studying 66 normal-hearing adults (41 men and 25 women) aged between 18 to 40 years who had been diagnosed with SCA (genotype HbSS) and were taking HDU medication. The episodes of crisis were found to be highly variable among all the participants. To limit the impact of this variable, all individuals needed to document a minimum of 5 crisis reports to be included. Based on their percentage HbF level at the time of the study, the participants were divided into three groups: Group 1 (10–15%), Group 2 (15–20%), and Group 3 (> 20%) (**Table 1**). Categorization was based on the observation that when the HbF level is 15 to 20%, health condition improves.

The inclusion criteria for the participants in the study were a prediagnosed SCA with HbF level < 30% tested within 3 months from the date of audiological evaluation, no prior otological issues, normal hearing sensitivity, and good health on the day of the experimental tests. Formal education up to the 5th standard and A-type tympanograms with reflexes were present in all subjects; their hearing thresholds were ≤15 dB HL at octave frequencies ranging from 250 to 8000 Hz. Additionally, it was determined through a structured interview that the subjects had no prior medical history of severe neurologic or otologic conditions. They did not receive payment for taking part in the study. The institute's ethics committee gave prior approval (ref. no. SH/EC/PhD/AUD-9/2023-24 dated 22-09-2023), and every participant gave written consent to voluntarily participate in the study.

There were two stages to the study. Based on the inclusion and exclusion criteria, phase I involved recruitment and grouping of participants. Phase II involved testing the chosen participants, including auditory discrimination tests and a speech perception in noise (SPIN) test.

Auditory discrimination tests (DLF, DLI, and DDT)

This involved three auditory tests for frequency, intensity, and duration discrimination. Difference limen for frequency (DLF), difference limen for intensity (DLI) [17], and a duration discrimination threshold (DDT) were done for 500 and 4000 Hz [18] tones at an anchor duration of 250 ms [19]. Stimuli were sampled at 44.1 kHz. The maximum likelihood procedure (MLP) toolbox in Matlab (MathWorks, USA) was used to evaluate all auditory discrimination tests [20]. Using a large number of candidate psychometric functions, the MLP determines the probability of receiving a response to each presented stimulus. The psychometric function yielding the highest probability determines the stimulus to be presented at the next trial. Within about 12 trials the MLP usually converges on a fairly stable approximation of the most likely psychometric function, which can then be used to estimate

Table 2. Median and IQR for auditory discrimination tests at 500 Hz and 4000 Hz, as well as SPIN at 0 dB SNR, across all three HbF groups

HbF level group		HbF 10–15%		HbF 15–20%		HbF > 20%	
Test parameter	**Test frequency**	**Median**	**IQR**	**Median**	**IQR**	**Median**	**IQR**
DLF	500 Hz	29.36	62.68	53.70	51.51	48.14	39.61
	4000 Hz	175.10	107.26	186.43	122.41	117.16	124.39
DLI	500 Hz	2.20	2.45	2.95	1.60	2.40	1.26
	4000 Hz	2.90	2.75	3.65	2.75	2.80	2.34
DDT	500 Hz	81.94	42.72	96.64	81.22	76.25	52.25
	4000 Hz	105.77	71.78	100.68	45.85	74.37	40.19
SPIN 0 dB SNR		31.0	6.0	34.5	5.0	34.0	5.0

Table 3. Kruskal–Wallis test statistic (*H*), significance level (*p*), degrees of freedom (*df*), auditory discrimination at 500 and 4000 Hz, and SPIN at 0 dB SNR across all three HbF groups

Test parameter	Test frequency	*H*	*df*	*p*-value
DLF	500 Hz	0.148	2	0.929
	4000 Hz	0.780	2	0.677
DLI	500 Hz	3.612	2	0.164
	4000 Hz	2.207	2	0.332
DDT	500 Hz	2.253	2	0.324
	4000 Hz	4.754	2	0.093
SPIN 0 dB SNR		1.477	2	0.478

threshold [21]. To track a 79.4% correct response criterion, an alternate forced choice method with three trials and MLP was utilized. Each trial consisted of three blocks with a stimulus presented in each; two blocks had the reference stimulus and the third block had the random variable stimulus. The task for the participants was to identify the blocks that held the variable stimulus. The aforementioned protocol was followed in each test, with the MLP toolbox was used to control stimulus presentation and response acquisition [22].

All psychoacoustical tests used a binaural test stimulus presented at 85 dB SPL; subjects were provided with 5 or 6 practice stimuli before the actual tests. A laptop (HP Intel Core i5) with Sennheiser HD449 headphones provided the stimuli for every test. The output of the headphones was adjusted to 85 dB SPL for pure tones at 500 and 4000 Hz in a 6 cc coupler.

Speech perception in noise (SPIN) test

For the SPIN test, the Hindi Sentence Test for Speech Recognition in Noise [23] at 0 dB SNR was utilized. To provide this level, each sentence was digitally combined with speech spectrum-shaped noise in Matlab. Using Sennheiser HD200A headphones, the stimuli plus noise were presented binaurally at 70 dB SPL (since the sentence list was initially standardized at this level [23]). There were 40 keywords in the sentence list. The target sentences needed to be written down or spoken aloud by the participants. The number of accurate keywords identified at 0 dB SNR was noted.

Results

The Statistical Package for the Social Sciences, SPSS (v.26) was used for statistical analyses of the data. A Shapiro–Wilk test showed that the data did not have a normal distribution in all three HbF groups for the three auditory discrimination tests (DLF, DLI, and DDT) at 500 or 4000 Hz or for the SPIN test at 0 dB SNR. Because of the data's non-normal distribution and large differences between mean and median values, median values of all parameters were utilized to compare groups. **Table 2** shows the median and inter-quartile range for DLF, DLI, DDT, and SPIN obtained at 0 dB SNR.

All the auditory discrimination tests were done at both 500 and 4000 Hz. The median and interquartile range (IQR) of all tests across HbF groups are depicted in **Table 2**. It can be seen that, for all auditory discrimination tests (DLF, DLI, and DDT), there is a trend for higher median

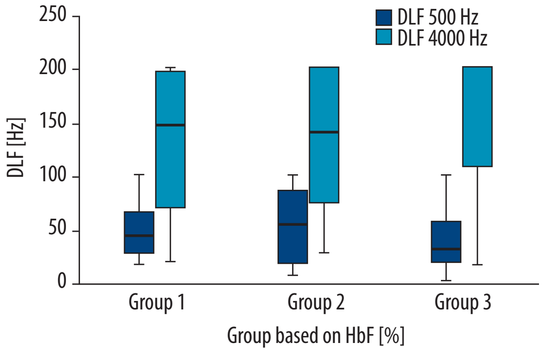

Figure 1. DLF at 500 and 4000 Hz for each of the three HbF groups. There is no statistically significant differences between the groups

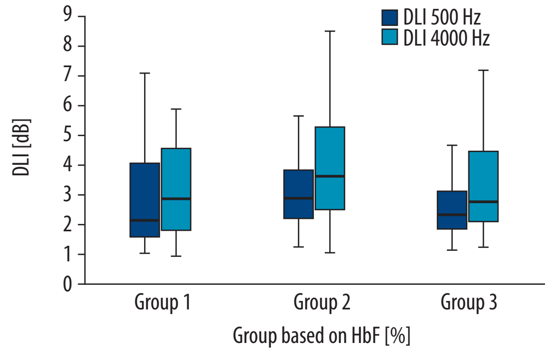

Figure 2. DLI at 500 and 4000 Hz for each of the three HbF groups. There is no statistically significant difference between the groups

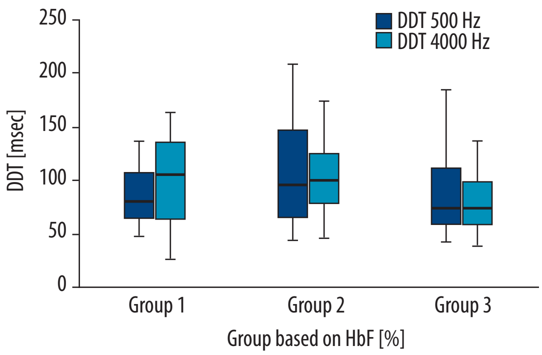

Figure 3. DDT at 500 and 4000 Hz for each of the three HbF groups. There is no statistically significant difference between the groups

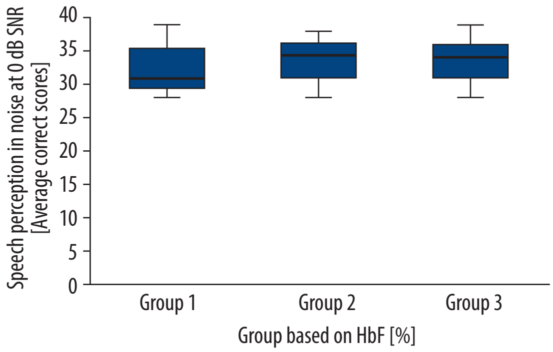

Figure 4. SPIN at 0 dB SNR for each of the three HbF groups. There is no statistically significant difference between the groups

and interquartile ranges with an increase in test frequency. Also, a slightly higher median (reflecting poorer responses) was observed for Group 2 with HbF% between 15–20%. Results of SPIN tests at 0 dB SNR showed poorer response for Group 1, having the lowest HbF% compared to the other two groups. For the SPIN test, the two higher HbF% groups had a similar median and IQR.

A Kruskal–Wallis test was run to assess the significance of differences between all HbF groups on the auditory tests (DLF, DLI, and DDT) in terms of the two tested frequencies (500 and 4000 Hz) and in terms of SPIN at 0 dB SNR (**Table 3**).

The results are shown graphically in **Figures 1, 2, 3, and 4**. There is no significant difference in scores among all three HbF groups in the auditory discrimination tests for each of the tested frequencies and similarly for SPIN scores. Since none of the test parameters were found to have significant differences among groups, correlations (between-group comparisons) were not computed.

Discussion

Individuals suffering from SCA sometimes report adverse effects on their auditory system, perhaps due to vaso-occlusion and hemolysis pathophysiology caused by stasis of the labyrinthine artery supplying the inner ear. This can involve hypoxia, ischemia, and organ cell death, leading to permanent inner ear damage and hearing loss [14] together with poor auditory processing ability and speech perception [15]. Ischemia of the highly metabolic cochlea and organ of Corti results in decreased oxygenation of the stria; vaso-occlusion can also contribute to labyrinthine hemorrhage and labyrinthitis ossificans causing sensorineural hearing loss [24].

SCA individuals who are under treatment with hydroxyurea (HDU) show varying levels of HbF. HDU is reported to be beneficial in preventing vaso-occlusive crises, probably by reducing the kinetics of HbSS polymerization and raising the volume of RBCs (reducing their deformability and improving the flow of blood through capillaries) and altering the expression of adhesion molecules [10].

In the present study, no significant effect was noticed across the three groups in any auditory processing test. This could be due to clinical absence or negligible raised HbF levels at the cochlear level, since unequal distributions of HbF across different organ systems has been reported [2]. It might also indicate no beneficial impact of raised HbF level upon the pathophysiological process of vaso-occlusion [12], at least at the level of the cochlea.

Further, though there were differences in HbF% across groups no significant difference suggests that even the HbF level below 15% and above 10% may be sufficient to increase blood flow. Data on HbF% before the assessment was not acquired, so this limits the study's conclusion and suggests further longitudinal investigation.

For all auditory discrimination tests, the high-frequency test frequency was found to give larger figures than the low-test frequency, but there was still no significant difference across HbF groups. The higher value with the 4000 Hz test frequency for discrimination tasks may be due to the asymmetrical anatomical distribution of blood supply throughout the length of the cochlea [25].

The SPIN scores were found to be slightly poorer for Group 1, although they were not statistically significant. Group 1 had HbF levels below the reported clinically beneficial level (i.e., < 15%). In the present study, participants were under HDU medication to boost HbF levels, and all had HbF levels between 10 and 30%, similar to previous findings [26]. The literature suggests a HbF level of > 15% is beneficial in reducing the overall impact of SCA-related pathophysiology. Although Groups 2 and 3 had HbF levels ≥ 15%, there was still no significant difference to the other groups; this probably supports the idea that even an HbF level of 10–15% may be sufficient to increase blood flow and provide similar functioning of the auditory system. This suggests that varying HbF levels did not change the functioning of the auditory system (either auditory discrimination or speech perception in noise).

Possibly, differences in speech perception seen in people with SCA may be related to the number of crises they have suffered. A larger number of crises might lead to poorer auditory processing difficulties or speech perception issues. In the present study, participants reported a large variability in the total number of crises, so this issue can be seen as one requiring further research. Finally, the timing of the tests administered to these individuals might be important, and this aspect also requires further investigation.

Conclusions

HbF levels of up to 15–20% have been reported to reduce clinical symptoms such as vaso-occlusion and hemolytic processes in SCA patients. The present study has found a negligible impact of raised HbF levels on auditory acuity. The explanation could involve unequal HbF distribution, negligible impact of improved HbF levels, and in general no direct link of HbF level with auditory performance. It seems that in SCA individuals every system in the body does not benefit equally or show a difference with raised HbF levels. Since the number of crises, which was highly variable among the participants, might play a major role, the outcomes of this study may not reflect the HbF level and its impact on auditory processing and speech perception outcomes.

Clinical implications

- The present study underlines the need for better understanding of the link between HbF level and the auditory system.
- There is a need for further studies on the impact of HDU and dose levels in SCA individuals to avoid its side effects on different organ systems, including the auditory system.
- Studying different auditory processing abilities in these individuals will give a better insight into microcirculatory changes in the cochlear and retro-cochlear regions after treatment.
- Choosing appropriate rehabilitative options for SCA individuals is still difficult.

Limitations and future directions

The present study evaluated the effect of HbF level on auditory processing at a specific point in time. Longitudinal studies might be the more effective way to understand the consequence of HDU treatment and HbF level in SCA individuals. Another limitation is the inability to measure the HbF level at the time of experimental test administration. Having knowledge of HbF levels at the same time as other variables like frequency of vaso-occlusive crises will give a better insight into impacts on the auditory system. Case controlled studies should be conducted in the future.

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F Literature analysis/search
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DOES IMPEDED BIOMECHANICS INFLUENCE COCHLEAR IMPLANT HEARING PRESERVATION?

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Abstract

Introduction: The insertion of a cochlear implant electrode may impede propagation of a travelling wave in the implanted cochlea. The primary objective of this study was to evaluate whether the impeded cochlea biomechanics affected the patient's hearing preservation.

Material and methods: There were 17 adults who were implanted with Flex 20 ($n = 1$); Flex 24 ($n = 8$); Flex 28 ($n = 1$); FlexSoft ($n = 5$); Medium ($n = 1$), and Standard ($n = 1$) electrode arrays with Pulsar, Concerto, or Sonata cochlear implants (Med-El). Implantation was via the round window. Acoustically evoked intracochlear potentials were recorded from cochlear implant electrodes. Tone pips at 500 Hz generated by a Nicolet EDX system (Natus Medical Inc.) were presented via inserts. Postoperative CT scans of the implanted cochlea were made and analysed. The unaided pre-implantation and post-implantation hearing thresholds were compared in all subjects.

Results: In 9 subjects the highest amplitude response to the tone pip matched the 500 Hz excitation area in the postoperative CT scan. The low-frequency difference was found between the pre-implantation and the post-implantation unaided thresholds in this group was 12.4 dB. Impeded basilar membrane biomechanics was observed in 8 other patients. In these 8 patients, the highest amplitude at 500 Hz was shifted either apically (1 case) or basally (7 cases) with respect to the 500 Hz excitation area suggested by the postoperative CT scan. The threshold difference in this group was 8.4 dB. However, the threshold differences in both groups were not statistically significant.

Conclusions: These preliminary data suggest that impeded biomechanics of the basilar membrane does not appear to affect hearing preservation.

Keywords: hearing preservation • cochlear implant • basilar membrane fixation

CZY ZABURZONA BIOMECHANIKA WPŁYWA NA ZACHOWANIE SŁUCHU PO WSZCZEPIENIU IMPLANTU ŚLIMAKOWEGO?

Streszczenie

Wprowadzenie: Głównym celem badania była ocena, czy zaburzona biomechanika ślimaka wpływa na zachowanie słuchu po wszczepieniu implantu ślimakowego.

Materiał i metoda: Siedemnastu dorosłym osobom wszczepiono implanty z elektrodą Flex 20 ($n = 1$); Flex 24 ($n = 8$); Flex 28 ($n = 1$); Flex Soft ($n = 5$); Medium ($n = 1$) i Standard ($n = 1$) z implantami ślimakowymi Pulsar, Concerto lub Sonata, Med-El Corp. Każdemu z badanych wszczepiono elektrodę implantu przez okienko okrągłe. Wewnątrzślimakowe akustyczne potencjały wywołane zostały zarejestrowane z elektrod implantu ślimakowego. Ton o częstotliwości 500 Hz był generowany przez system Nicolet EDX, Natus Corp i podawany przez słuchawki wewnątrzuszne. Po operacji wykonano tomografię komputerową. Audiogram przed implantacją porównano z audiogramem wykonanym podczas badania.

Wyniki: Dziewięciu pacjentów miało najwyższą amplitudę odpowiedzi na ton 500 Hz na elektrodzie odpowiadającej obszarowi wzbudzenia dla tonu 500 Hz określonemu na podstawie pooperacyjnej tomografii komputerowej. Zaburzoną biomechanikę błony podstawnej zaobserwowano u 8 pacjentów. U tych pacjentów najwyższa amplituda w odpowiedzi na ton o częstotliwości 500 Hz była przesunięta w kierunku szczytowym (1 przypadek) lub podstawnym (7 przypadków) w stosunku do obszaru pobudzenia o częstotliwości 500 Hz ocenianego w pooperacyjnej tomografii komputerowej. Średni spadek czułości słuchu mierzony w audiometrii tonalnej w grupie osób z tonotopią wynosił 12,4 dB, podczas gdy w grupie z zaburzoną biomechaniką wynosił 8,4 dB. Różnica ta nie była istotna statystycznie.

Wnioski: Wstępne dane sugerują, że zaburzona biomechanika błony podstawnej nie ma wpływu na zachowanie słuchu.

Słowa kluczowe: zachowanie słuchu • implant ślimakowy • biomechanika błony podstawnej

Introduction

Hearing preservation after cochlear implantation is considered the most important factor in rating the success of the procedure [1]. The risk of causing trauma to the cochlea when implanting an electrode array is real, and could ultimately lead to complete hearing loss. Although relatively good hearing-preservation rates are achieved using lateral-wall flex electrodes [2], there is still a need to improve hearing-preservation rates.

One of the ways to assess hearing preservation is to look for evidence of a travelling wave inside the cochlea. Ever since 1935 when Fromm and colleagues first measured the cochlear potential in three subjects, electrocochleography (ECochG) has become a reliable measure of cochlear function. More recently, intracochlear ECochG has been introduced, and its feasibility was presented at the XXXII World Congress of Audiology in 2014 [3].

When pure-tone stimuli are acoustically applied to the ear, they generate a mechanical wave in the basilar membrane that progresses from the cochlear base and peaks at a place that corresponds to the frequency of the tone. After passing this region of the cochlea, the amplitude of the wave drops. This phenomenon was discovered and first measured in human-ear specimens by von Békésy in 1928 [4]. In the case of basilar-membrane fixation caused by electrode implantation, the wave energy is focused in regions adjacent to the point of fixation [5]. The impeded biomechanics of the basilar membrane not only affects the amplitude of the traveling wave, but also its phase.

The goal of this study is to evaluate whether impeded basilar-membrane biomechanics affects the hearing preservation in patients implanted with flex electrode arrays.

Material and methods

The Ethics Committee of the Institute of Physiology and Pathology of Hearing (decision KB.IFPS 16/2021) approved the study protocol. Prospective subjects were given an informed consent form that explained the purpose and procedures involved. The test procedures were in accordance with the ethical standards of the Helsinki Declaration.

Seventeen adults (9 females and 8 males) were selected for the study who had varying degrees of hearing abilities, both before and after receiving their cochlear implants (CIs). The subjects were implanted with the following electrode arrays: Flex 20 ($n = 1$), Flex 24 ($n = 8$), Flex 28 ($n = 1$), Flex Soft ($n = 5$), Medium ($n = 1$), and Standard ($n = 1$) (all Med-El) of Pulsar, Concerto, or Sonata cochlear implants (Med-El). Implantation was done using the round window insertion technique [6]. The mean age was 47 years and 2 months at the time of surgery (20–68 years). The mean time of measurements was 13 months (2–91 months after CI surgery). To minimise the damaging effect of the electrode insertion and to make hearing preservation possible, we selected electrodes suitable for deep insertion, i.e., reaching the region of 500 Hz and below (i.e., 250 Hz).

Recordings of intracochlear ECochG

Intracochlear acoustically evoked potentials were recorded from the CI electrodes. Patients were positioned in a comfortable semi-lying position. Inserts were placed inside their implanted ear. The inserts were connected to the Nicolet EDX system (Natus Medical Inc.), which was used for acoustic stimulation. The Meastro AP research software (Med-El), was run from a PC communicating with a MAX Interface (Med-El). The MAX interface communicated via an external coil connected to the CI, which is possible when the coil is placed on the CI. When recording was initiated, the MAX interface triggered the Nicolet EDX, which acoustically stimulated the patient. TIP 300 inserts were used during the acoustic stimulation.

Short 8 ms (4 cycle) 500 Hz tone pips were used as acoustic stimuli. The stimuli had condensation polarity. Before measuring the intracochlear ECochG, the subject's maximum comfortable level (MCL) was obtained. All recordings were then performed at the MCL. The mean stimulation level was 103.8 dB HL (85–122 dB HL). For each subject, an unaided preoperative audiogram was measured less than 4 weeks prior to surgery, and an unaided postoperative audiogram was measured on the day of the study. For situations where no acoustic threshold was obtained at 115 dB HL, 120 dB HL was used. At the time of the study, a postoperative CT scan was obtained for each subject and the electrode placement was evaluated. Further details on the technical procedure, as well as other tests performed to evaluate possible artefacts, see [7].

The location of the intracochlear electrode contacts was derived from the postoperative computed tomographic imaging. The Greenwood function was used to infer the characteristic frequency corresponding to the electrode location [8,9]. Estimation of the cochlear angle of rotation and characteristic frequency region was done according to Polak et al. [10], which has the characteristic location for 500 Hz at 457°. In all patients, the electrode placement that was closest to the insertion angle of 457° was estimated to correspond to the 500 Hz excitation area.

After the intracochlear ECochG was recorded at all electrode contacts, the 500 Hz frequency place was estimated from the response, and the electrode with the maximum amplitude was selected. If the maximum amplitude was registered within one contact of the selected tonotopic electrode identified in the postoperative CT scan,

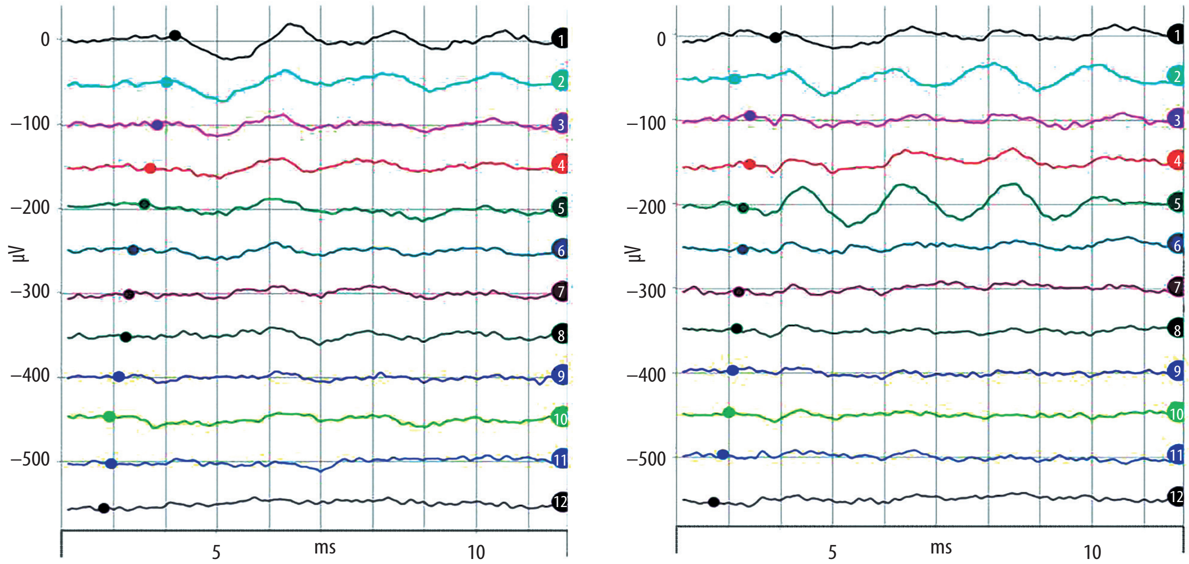

Figure 1. Examples of intracochlear ECochG recordings of 500 Hz tone pips at 95 dB HL from all 12 electrodes of the Med-El Flex 24 array, as captured by the Maestro AS software. *Left*: Examples from a patient in the tonotopy group. The electrode insertion depth was 450°. From the postoperative CT scan, the expected maximum amplitude was at electrode 1 (8.2 µV), as confirmed by the intracochlear ECochG electrode 1. *Right*: Example of an ECochG recording of a patient from the non-tonotopy group. The electrode insertion depth was 520°. From the postoperative CT scan, the expected maximum amplitude should be at electrode 2. However, the intracochlear ECochG showed the largest response at electrode 5 (17.5 µV). An additional local minimum was found at electrode 2 (8.9 µV). Electrode 1 is the most apical and electrode 12 the most basal. The coloured dots mark the latencies of the responses

the patient was classified as belonging to the tonotopy group. Otherwise, the patient was classified as belonging to the non-tonotopy group.

Data analysis

To compare the postoperative audiometric thresholds and the differences between preoperative and postoperative thresholds for all characteristic frequencies between the tonotopy and non-tonotopy groups, a two-tailed *t*-test was used. To evaluate if the data was normally distributed, a Lilliefors test of conformity was performed, using a significance criterion of $\alpha = 0.05$. The sample size was selected to achieve statistical power greater than 0.9.

Results

The mean electrode insertion at the tip of the array was 515° (381–729°). In 16 out of 17 cases the excitation area of 500 Hz was reached. The tonotopy group comprised 9 patients and the non-tonotopy group comprised 8 patients.

Figure 1 shows an example of intracochlear ECochG recordings of 500 Hz tone pips in patients from both the tonotopy and the non-tonotopy groups. In the tonotopy group recording, the maximum amplitude matched the electrode number estimated from the postoperative CT scan. In contrast, in the non-tonotopy-group, the difference between the maximum amplitude electrode and the estimated electrode number from the postoperative CT scan were two contacts or more. **Figure 1** (right) shows that the travelling-wave energy is focused between electrodes 5 and 2, suggesting that the point of fixation lies between them. In 7 other cases, the maximum amplitude was more basal and in one case it was more apical. For the tonotopy group, the 500 Hz peak was at 442° (360–540°), which was consistent with the Polak et al. model estimate of 457°. However, for the non-tonotopy group, the mean peak angle was 170° (45–360°).

Figure 2 depicts the mean and standard deviation of the unaided postoperative audiograms of the tonotopy and non-tonotopy groups. The difference between the postoperative and preoperative mean threshold are plotted as well. For the tonotopy group, the low-frequency (125–1000 Hz) pure-tone average difference was 12.4 dB, while for the non-tonotopy group it was 8.4 dB.

The hearing thresholds of both test groups were normally distributed at all frequencies, according to the Lilliefors test of normality (using a significance level of 0.05 and *p*-value lower than the critical value $Dn = 0.285$ for all frequencies). No statistical differences between preoperative and postoperative thresholds were found in any of the groups (two-tailed *t*-test: $\alpha = 0.05$, $p > 0.05$). **Table 1** provides the numerical threshold data of **Figure 2**.

Discussion

This study evaluated hearing preservation in patients implanted with various Med-El Flex electrodes. One of the measures of hearing preservation is evidence of a travelling wave in the cochlea. The study found that after electrode implantation not all patients exhibited propagation of a travelling wave induced by 500 Hz tone pips. In the non-tonotopy group of patients, the measured

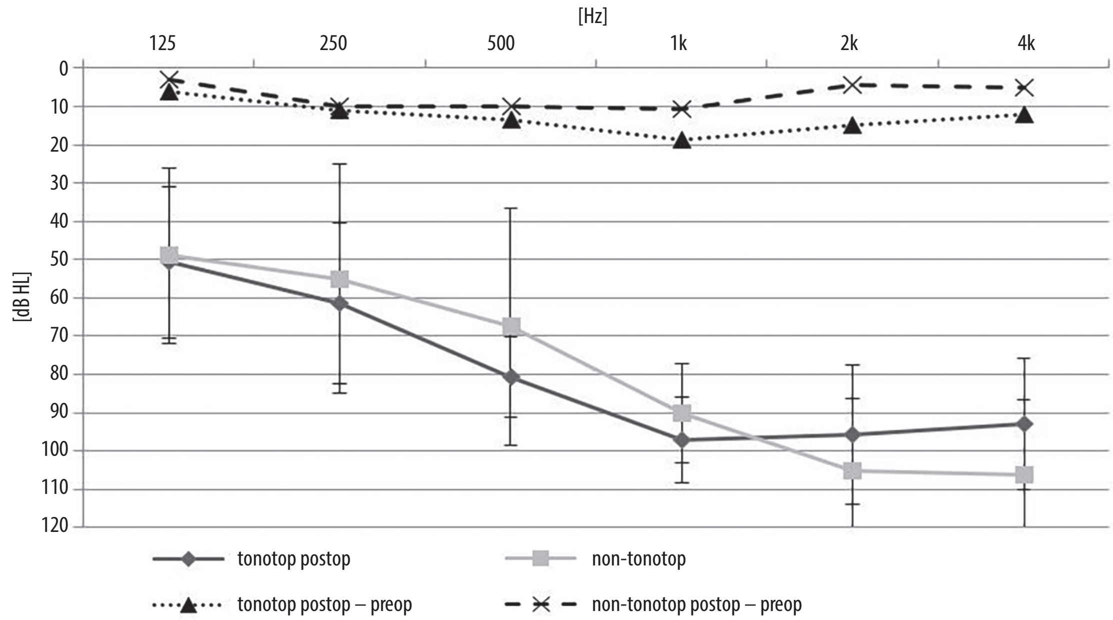

Figure 2. Mean unaided postoperative audiograms of the tonotopy and non-tonotopy groups (continuous lines) and the mean difference between their preoperative and the postoperative audiograms (dotted lines)

Table 1. The numerical data of the plots in Figure 2

Frequency	125 Hz	250 Hz	500 Hz	1 kHz	2 kHz	4 kHz
Tonotopy group postop mean ± *SD* [dB HL]	48.9 ± 22.9	55 ± 29.9	67.5 ± 31.1	90 ± 12.9	105 ± 18.7	106.3 ± 19.5
Non-tonotopy group postop mean ± *SD* [dB HL]	50.7 ± 19.9	61.4 ± 21	80.7 ± 10.6	97.1 ± 11.1	95.7 ± 18.1	92.9 ± 17
Tonotopy group postop – preop mean ± *SD* [dB HL]	6.1 ± 17.6	11.1 ± 17.5	13.3 ± 12.2	18.9 ± 20.1	15 ± 7.9	12.2 ± 10.3
Non-tonotopy group postop – preop mean ± *SD* [dB HL]	2.9 ± 14.1	10 ± 13.5	10 ± 12.6	10.7 ± 9.3	4.3 ± 3.5	5 ± 8.2

maximum amplitude in the electrode recordings did not match the expected excitation area of 500 Hz according to the Greenwood and Polak models. The sound energy in the cochleas of these patients was concentrated in regions adjacent to the point of basilar membrane fixation [5].

Eshraghi et al. [11] proposed a grading system for cochlear trauma ranging from 0 to 4, where a higher grade indicates greater cochlear trauma. Grade 0 is defined as no trauma, while Grade 1 corresponds to a mechanical elevation of the basilar membrane. In our study, we identified 8 patients with impeded biomechanics of the basilar membrane. However, we found that this group did not experience poorer hearing preservation 1 year after cochlear implantation compared to the group without any impeded biomechanics. This finding suggests that touching or elevating the basilar membrane to only a certain degree does not significantly affect the hearing loss induced by cochlear implantation and may, therefore, still be considered as Grade 0.

Bester et al. [12] evaluated 39 patients implanted with Cochlear Nucleus CI422 and CI522 CIs with a 20 mm-long straight electrode array with 22 contacts. The electrode insertions varied between 20 to 25 mm. Tone pips of 500 Hz were applied and recorded at all 22 contacts along the electrode array. In perioperative ECochG recordings, 22 subjects (56%) had the peak response at around the tip of the electrode array (apical-peak, AP; EL20 or EL22), whereas 17 subjects (44%) had maximum amplitude in more basal regions (mid-peak, MP; EL18 or lower). The responses were not compared with postoperative CT scans. In 6 subjects with perioperative apical peaks, the location of the largest ECochG response had shifted basally (apical-to-mid-peak, AP–MP). The mean postoperative hearing loss in the AP group was 13 dB ($n = 16$, $SD = 9$). A significantly larger hearing loss was detectable in the MP and AP–MP groups with 28 ($n = 17$, $SD = 10$) and 35 dB ($n = 6$, $SD = 13$). The authors concluded that after cochlear implantation, MP and AP–MP ECochG response patterns were correlated with poorer postoperative hearing and higher four-point impedances in comparison with AP response patterns, all of which may suggest increased intracochlear fibrosis.

In our study, we did not find any difference in the hearing preservation between the tonotopic and the non-tonotopic groups. The differences may be in the type of array used and in the electrode insertion depth. In our study, the mean insertion depth was 515° (381–729°) and the 500 Hz excitation area was reached in all but one case, whereas the mean electrode insertion achieved with the CI422/CI522 slim straight electrode was 373° (233–470° [13]), which suggests that the insertion depth was always equal to or more basal than the 500 Hz excitation area. Therefore, the most apical electrode contact in all cases is the one closest to the excitation area of 500 Hz.

Walia et al. [14] evaluated 50 adults implanted with the Cochlear CI632 perimodiolar array, with a mean electrode insertion depth of 404° ± 35°. Tone burst stimuli were independently administered at frequencies of 250, 500, 1000, 2000, 3000, and 4000 Hz for all 22 electrodes, and intracochlear ECochG recordings were obtained. For each frequency presented, the electrode with the highest peak was selected as the excitation frequency area. Based on this information, the authors created individual tonotopic maps for frequencies between 500 and 4000 Hz. The authors found a near-octave apical shift from the Greenwood model at all frequencies. In our case, for a 500 Hz tone burst, the mean peak place was 295° (45–630°), while the mean 500 Hz peak reported by Walia et al. was 330° (220–380°). The similarity in results between both studies may be explained by impeded cochlear biomechanics. In our study, for the tonotopy group the mean peak place was 442° (360–540°), which was consistent with the Greenwood and Polak et al. models (estimated to be 457° at 500 Hz). However, the mean insertion angle at the peak was 170° (45–360°) in the non-tonotopy group.

Conclusions

Our preliminary data suggest that impeded biomechanics of the basilar membrane does not necessarily impair hearing preservation. It would be of interest to investigate this phenomenon in more detail and specifically find out whether basilar-membrane impedance of the degree that was found here has any negative effect on patients.

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EFFICACY OF SMARTPHONE-BASED SCREENING BY TEACHERS FOR EARLY IDENTIFICATION OF HEARING LOSS IN SCHOOL CHILDREN

Contributions:
A Study design/planning
B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

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Abstract

Introduction: The high prevalence of hearing impairment in school children highlights the need for regular school screening programs. However, lack of professionals and infrastructure can create a barrier for screening all children. Smartphones present an opportunity for school teachers to be trained to perform hearing screening among students. The current study aimed to determine the efficacy of smartphone-based hearing screening by school teachers.

Material and methods: The participants comprised 580 students, from grades 1 through 5 from various schools in Dharwad city. The children were screened by trained school teachers using the smartphone-based app Hearing Test developed by e-audiologia.pl. To test validity, the same children were again tested by a professional audiologist using a diagnostic clinical audiometer.

Results: The results of the current study found small but significantly higher mean thresholds across frequencies using the Hearing Test app compared to the diagnostic audiometer. However, thresholds obtained using both devices were within normal limits of –10 to 15 dB HL. Hence, it might be possible for the Hearing Test app to be used for hearing screening in primary school children.

Conclusions: After training, it appears feasible for school teachers to utilize the Hearing Test app to screen for hearing loss in school children. This could make hearing screening more routine and cost-effective, and may aid in the early detection of hearing loss. However, it appears that hearing thresholds established by teachers using the app were slightly worse than those established by an audiologist using an audiometer.

Keywords: school children • hearing screening • Hearing Test app • audiometry • hearing threshold

SKUTECZNOŚĆ BADAŃ PRZESIEWOWYCH WYKONYWANYCH PRZEZ NAUCZYCIELI ZA POMOCĄ SMARTFONÓW W CELU WCZESNEJ IDENTYFIKACJI UBYTKÓW SŁUCHU U DZIECI W WIEKU SZKOLNYM

Streszczenie

Wprowadzenie: Wysoka częstość występowania zaburzeń słuchu u dzieci w wieku szkolnym implikuje potrzebę prowadzenia regularnych programów badań przesiewowych w szkołach. Jednak brak specjalistów i odpowiedniej infrastruktury może stanowić przeszkodę wykonywania badań przesiewowych u wszystkich dzieci. Wykorzystanie aplikacji na smartfony stwarza możliwość przeprowadzania w szkołach – po przeszkoleniu nauczycieli w tym zakresie – badań przesiewowych słuchu wśród uczniów. Niniejsze badanie miało na celu określenie skuteczności wykonywanych w szkołach przez nauczycieli badań przesiewowych słuchu za pomocą smartfonów.

Materiał i metody: W badaniu wzięło udział 580 uczniów z klas od 1 do 5 z różnych szkół w mieście Dharwad. Dzieci zostały przebadane przez przeszkolonych nauczycieli za pomocą aplikacji na smartfony Hearing Test opracowanej przez e-audiologia.pl. Aby sprawdzić poprawność wyników, te same dzieci zostały ponownie zbadane przez profesjonalnego audiologa z wykorzystaniem audiometru klinicznego.

Wyniki: Wyniki badania wykazały niewielkie, ale znacząco wyższe średnie progi słyszenia dla różnych częstotliwości uzyskane w aplikacji Hearing Test w porównaniu z audiometrem klinicznym. Jednak progi uzyskane za pomocą obu urządzeń mieściły się w granicach normy od –10 do 15 dB HL. Wynika stąd, że aplikacja Hearing Test może być wykorzystywana do badań przesiewowych słuchu u dzieci w wieku szkolnym.

Wnioski: Wydaje się możliwe, aby po przejściu szkolenia nauczyciele w szkołach korzystali z aplikacji Hearing Test do wykonywania badań przesiewowych w kierunku ubytku słuchu u dzieci w wieku szkolnym. Tym samym badania przesiewowe słuchu mogłyby stać się bardziej powszechne i przystępne cenowo, a dzięki temu mogłyby pomóc we wczesnym wykrywaniu ubytków słuchu. Jednocześnie należy wziąć pod uwagę, że progi słuchu ustalone przez nauczycieli w niniejszym badaniu były nieco gorsze niż te ustalone przez audiologa za pomocą audiometru klinicznego.

Słowa kluczowe: dzieci w wieku szkolnym • badania przesiewowe słuchu • aplikacja Hearing Test • audiometria • próg słyszenia

Introduction

If left untreated, communication disorders in childhood can result in severe consequences, such as limited educational achievement, reduced employment opportunities, and problems with social adaptation. We now have ample evidence that shows early identification of communication disorders reduces its impact on social, emotional, and educational outcomes [1–3]. A review of the literature from 1980 to 2020 showed that the prevalence of hearing loss in children in India ranged from 6 to 27% [4]. Another survey conducted in a rural population of India found that the prevalence of individuals at risk of communication disorders was 6.1%; among those at risk, the prevalence of audiological and or otological disorders was found to be 90.6% [5]. Similarly, in a retrospective analysis of clinical records of individuals having communication disorders, hearing impairment reached a prevalence of 30.8% in children [6].

A prevalence study in the Netherlands reported 7.8% of children 9–11 years old had sensorineural hearing loss in one or both ears, and that a history of recurrent acute otitis media and low maternal education were the most common predisposing factors for hearing loss [7]. A hearing screening in 67,416 school children from rural areas in Poland found positive results in 16.4%; untreated middle ear diseases were associated with a higher prevalence of hearing loss in rural areas [8]. Among 34,618 elementary school children screened in Poland, 11% had sensorineural hearing loss [9]. It is clear that in preschool and school age children the prevalence of communication disorders can be high, and it can often go undetected if they are not screened.

School screening typically uses a systematic approach involving otoscopy and pure-tone audiometry to examine a large number of children from a large geographical area [8]. In the Polish work, screening was carried out in an isolated room having low noise background, and results were considered normal if the child had air conduction hearing thresholds less than 20 dB HL [10,11]. Although audiological screening requires minimum equipment, costs involved in purchasing and maintaining it can be high. Smartphone apps are inexpensive, readily available, and easy to use. Several research groups have looked at hearing testing using a smart phone [12–15]. Smartphone apps have the potential to be a convenient alternative to employing professionals in screening children's hearing. Smartphone apps for basic hearing assessment might provide low-cost hearing screening at the Primary Health Centre (PHC) level [16].

There is growing evidence that smartphone-based hearing screening can be effective in identifying hearing loss. A systematic review and meta-analysis found that smartphone-based hearing tests have a sensitivity ranging from 0.71 to 1.00 and specificity ranging from 0.73 to 1.00 when compared to conventional audiometry. One study found that smartphone-based hearing tests were reliable and accurate across different smartphone models and operating systems [13]. Among adults, smartphone based self-test audiometry provided accurate and reliable results having a sensitivity of 90.6% [17]. Similarly, the sensitivity and specificity of a smartphone-based hearing screening app hearScreen were 81.7% and 83.1%, respectively; positive and negative predictive values of 87.6% and 75.6%, respectively, have also been reported [18]. Likewise, validity of the Hearing Test app revealed comparable sensitivity (75.0%) and specificity (98.5%) compared with conventional screening audiometry [19].

Here, the validity of hearing screening using Hearing Test smartphone-based audiometry is explored. In the self-test response mode, hearing screening with the similar hearTest app appears to be reliable in detecting hearing loss in adults and children with hearing loss [20]. These findings suggest that smartphone-based hearing screening might be a useful tool for identifying hearing loss, particularly in low-resource settings where access to traditional audiological services may be limited.

In school children, hearing screening is important to identify late onset hearing loss, unilateral hearing loss, or cases missed at newborn hearing screening. In India, hearing screening in school children is not universal due to the lack of professionals and infrastructure. The use of a smartphone app for hearing screening is promising as it requires low-cost instrumentation and minimal training. This study investigates the effectiveness of smartphone-based hearing screening by teachers of primary school children. We wanted to see whether teachers could be trained to use a smartphone app to obtain hearing thresholds in these children.

Material and methods

Participants

Primary school children aged between 5 to 10 years (mean age 7.2, *SD* 2.3 years) and studying in grades 1 to 5 were selected through a simple random sampling method. There were 580 (317 male, 263 female) participants in the study, as shown in **Table 1**. The participants had no symptoms related to hearing loss. There were 79 other students who had a history of ear pain, ear discharge, or difficulty in following instructions; data from them were analyzed separately for a subsequent study.

Methods

For hearing screening the Android-based app Hearing Test (developed by e-audiologia.pl), available at no cost in the Google play store, was used. Compared to other apps, Hearing Test has a lower cutoff threshold (20 dB HL); further, it has good sensitivity (98%) and specificity (79%) and

Table 1. Distributions of students in terms of class and gender

	Class				
	I	II	III	IV	V
Gender					
Male	64	66	61	66	55
Female	32	35	44	43	35
Total	96	101	105	109	90

has been recommended for large-scale screening and epidemiological studies [15]. The app produces pure tones from 0.125 to 10 kHz at intensities ranging from 0 to 100 dB HL. Two different headphone options were available: a bundled headphone, in which calibration is automatically provided in the app, and an unbundled headphone which requires manual calibration prior to test commencement. In our study, teachers used unbundled headphones (Fingers Superstar H6) and calibration was performed before each session. Calibration was done on a young graduate having normal hearing sensitivity in both ears. The subject was asked to self-record his hearing thresholds by pressing or releasing the button as the intensity changed. Frequency was slowly increased from 125 Hz to 10,000 Hz at 1 octave per minute and 2 dB per second. The thresholds obtained during calibration were verified using a clinical audiometer. An Android v. 11.0 smartphone (Redmi note 10S) was used for the entire data collection. The clinical audiometer was an ALPS AD 2000 with TDH 39 headphones that were calibrated as per ANSI S3.6-2018 (R2023). The audiometer produced pure tones ranging from 0.25 to 8 kHz at intensities from –10 to 120 dB HL for air-conduction. Using the two devices, hearing thresholds could be established at 0.5, 1, 2, and 4 kHz.

Procedure

The study was conducted at various primary schools in Dharwad, North Karnataka, India. Informed consent was collected from the parents before the scheduled date of the hearing screening. The study was carried out in three phases (**Figure 1**), and adhered to the ethical guidelines of the institution (JSSMC/IEC/070326/02NCT/2023-24 dated 9.03.2024). In phase 1, 15 teachers were enrolled in a training program for a week and were provided with information on the screening process and hands-on training using the smartphone-based hearing app. They were trained by an experienced audiologist who outlined the importance of hearing, different types of hearing loss, and early identification of hearing loss. Training was done for an hour per day continuously for a week. Every teacher was given a manual in Kannada, describing step-by-step instructions on how to calibrate the headphones prior to the testing, how to modify the stimulus frequency and intensity, and how to save and retrieve test data. At the end of training, they were asked to answer a post-test containing simple questions related to hearing loss and its early identification; they were also asked to perform hearing screening using the mobile app. Six teachers who completed the post-test and hearing screening with 80% accuracy were chosen for further study. In phase 2, the 6 chosen teachers administered smartphone-based hearing screening for grade I to V students. Phase 3 involved use of the clinical audiometer, and conventional audiometric thresholds were established by an experienced clinical audiologist. The interval between the last two phases was less than one week. Phases 2 and 3 were carried out in a quiet room in the school that had low background noise. The validity of the smartphone-based screening was done by comparing the two sets of results.

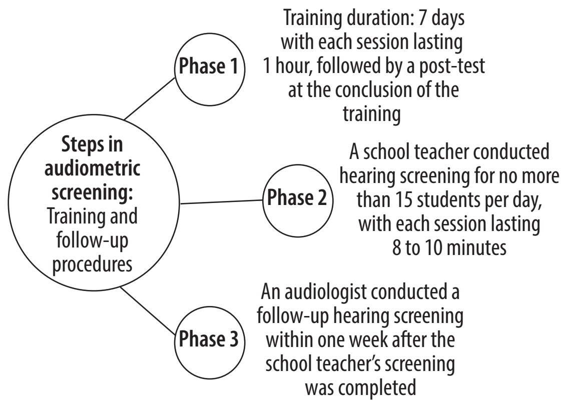

Figure 1. Steps involved in hearing screening

Results

The study compared hearing thresholds obtained using the Hearing Test app and a clinical diagnostic audiometer across four different frequencies (0.5, 1, 2, 4 kHz). The data was analyzed using Statistical Package for Social Sciences (SPSS), Windows version 21 software. The significance criterion was $p < 0.05$. The results were analyzed using mixed ANOVA where the testing devices served as within-subject independent variable, while class and gender served as between-subject variables. Hearing thresholds at a particular frequency served as the dependent variable.

Table 2 shows that the mean thresholds obtained using the Hearing Test app were slightly higher than that of the diagnostic clinical audiometer at all frequencies in all five classes. Mean thresholds obtained using the Hearing Test app ranged between 13–16 dB HL (SD 3–8 dB HL) across the four frequencies, whereas using conventional audiometry, the hearing thresholds ranged between 2–12 dB HL (SD 4–8 dB HL).

Table 3 shows the results of mixed ANOVA at all four frequencies, where a significant main effect of testing device ($p < 0.05$) and an interaction between testing device and

Table 2. Means and standard deviations across four frequencies of hearing thresholds [dB HL] obtained using the Hearing Test app and diagnostic clinical audiometer in children from 1st to 5th class

		Class									
		I		II		III		IV		V	
Frequency		**Aud**	**App**	**Aud**	**App**	**Aud**	**App**	**Aud**	**App**	**Aud**	**App**
500 Hz	Mean	5.39	14.32	10.71	13.14	9.90	13.47	11.46	14.40	7.58	13.69
	SD	5.05	3.54	7.00	4.11	8.48	4.55	7.11	4.22	6.61	4.29
1000 Hz	Mean	7.26	13.75	11.78	13.49	9.66	13.59	9.70	15.60	8.25	14.22
	SD	4.63	3.22	6.27	4.28	7.94	4.72	8.62	4.46	6.15	4.77
2000 Hz	Mean	9.06	14.11	14.38	14.00	11.64	13.78	8.30	14.52	12.16	14.47
	SD	5.53	3.18	4.65	4.09	4.78	5.12	6.10	4.37	4.45	4.87
4000 Hz	Mean	2.83	14.09	8.63	13.81	8.14	15.21	6.51	16.55	4.91	15.16
	SD	4.56	3.94	5.83	3.99	5.77	4.98	6.23	4.56	4.79	4.36

Note: Aud = clinical audiometer; App = Hearing Test app

Table 3. Results of mixed ANOVA showing significant interactions

	Variables			
	500 Hz	**1000 Hz**	**2000 Hz**	**4000 Hz**
Testing device and class	$F = 8.794$	$F = 4.838$	$F = 13.501$	$F = 10.203$
	$p < 0.0005$	**$p = 0.001$**	**$p < 0.0005$**	**$p < 0.0005$**
Testing device and gender	$F = 0.138$	$F = 0.127$	$F = 0.775$	$F = 0.058$
	$p = 0.710$	$p = 0.722$	$p = 0.379$	$p = 0.810$
Gender and class	$F = 0.390$	$F = 0.314$	$F = 0.976$	$F = 1.343$
	$p = 0.816$	$p = 0.869$	$p = 0.420$	$p = 0.253$
Testing device, gender, and class	$F = 1.567$	$F = 2.536$	$F = 2.958$	$F = 3.007$
	$p = 0.182$	**$p = 0.039$**	**$p = 0.020$**	**$p = 0.018$**

Note: bold = results statistically significant

class ($p < 0.05$) can be seen. Further, the interaction between testing device, gender, and class were also significant for 1, 2, and 4 kHz ($p < 0.05$). However, there was no interaction between testing device and gender ($p < 0.05$). Between subjects, the main effect of gender ($p > 0.05$) and class ($p > 0.05$) was not significant. Similarly, there was no interaction between gender and class ($p > 0.05$).

Since there was a significant interaction between testing device and class, hearing thresholds obtained using the two devices were compared in each class using a paired sample *t*-test. **Table 4** shows that at all the frequencies, hearing thresholds obtained using the diagnostic clinical audiometer were significantly better ($p < 0.05$) than the Hearing Test app in all 5 classes.

Discussion

The main objective of the study was to investigate the effectiveness of using a smartphone-based hearing app for screening of primary school-children by teachers. Comparison of hearing thresholds between the app and the diagnostic audiometer showed that smartphone-based screening gave significantly higher thresholds than the audiometer. The smartphone gave hearing test thresholds of 13–16 dB HL across frequencies from 0.5 to 4 kHz, whereas conventional audiometry gave hearing thresholds of 2–12 dB HL. Comparing classes, the thresholds for all five classes were similar. A previous study among adults which compared hearing thresholds using an audiometer and the Hearing Test app showed that hearing thresholds were 0–6.5 dB using the audiometer and 0–7.5 dB using the app [15]. The study noted that the low thresholds recorded from both devices were possible because all measurements were carried out in a sound booth. In contrast, we measured thresholds in a quiet room in the school having low background noise.

In the current study, the mean difference in hearing thresholds between audiometer and the Hearing Test app ranged from 2 to 12 dB (SD 1–4 dB). In comparison, the study by Masalski et al. [15] reported a mean difference of 2.6 (*SD* 8.3 dB). The significant difference in thresholds obtained using the two devices in the current study can be attributed to several reasons. The present study used unbundled headphones that were calibrated before every test, whereas the former study used bundled headphones that were calibrated specific to the smartphone manufacturer. In the

Table 4. Results across frequencies of paired sample *t*-tests comparing hearing thresholds obtained using the Hearing Test app and a diagnostic clinical audiometer in children from the 1st to 5th class

Frequency	*t*-test	Class I	Class II	Class III	Class IV	Class V
500 Hz	*t*-value	14.425	2.872	3.940	3.811	6.663
	df	95	100	104	108	89
	p	**< 0.0005**	**0.005**	**< 0.0005**	**< 0.0005**	**< 0.00005**
1000 Hz	*t*-value	11.087	2.256	4.179	6.065	7.397
	df	95	100	104	108	89
	p	**< 0.0005**	**0.026**	**< 0.0005**	**< 0.0005**	**< 0.0005**
2000 Hz	*t*-value	8.112	–0.610	3.063	8.105	3.924
	df	95	100	104	108	89
	p	**< 0.0005**	**0.543**	**0.003**	**< 0.0005**	**< 0.0005**
4000 Hz	*t*-value	20. 344	7.726	9.804	14.146	13.580
	df	95	100	104	108	89
	p	**< 0.0005**	**< 0.0005**	**v0.0005**	**< 0.0005**	**< 0.0005**

Note: *df* = degree of freedom; *p* = statistical significance; bold = results statistically significant

Masalski study, the tone used was 100% modulated, which is easy to detect in noise, whereas in the current study a steady pure tone was used. Further, in the Masalski study, the entire testing procedure was under the guidance of an audiologist whereas in the current study the entire testing was performed by a teacher. Another study of adults that compared hearing thresholds obtained using an audiometer and the Hearing Test app found mean threshold differences up to 8.8 dB [19].

Using the Hearing Test app, none of the children had hearing thresholds greater than 20 dB HL at any frequency, indicating that all hearing thresholds were within normal limits. In general, screening results are considered normal if a child has air conduction thresholds better than 20 dB HL [10,11,19]. Hence, in an Indian context, we conclude that the Hearing Test app can be used by teachers for hearing screening of school children.

The present study validates the use of smartphone-based hearing screening in school children between grades 1 to 5. Most researchers have studied the specificity and sensitivity of app-based identification of hearing loss in adults [13–17]. However, a few studies have compared conventional screening audiometry and smartphone-based screening in normal and hearing-impaired in children as well as adults [18,20]. The findings of the current study reinforce the appropriateness of smartphone-based hearing screening among primary school children.

In the current study, comparisons were made in both males and females, and both had similar hearing thresholds within the normal limits. There was no gender difference across frequencies, testing devices, and classes, supporting earlier reports [21,22]. It may be concluded from the current study that some school teachers can be trained to use the smartphone-based Hearing Test app for hearing screening of school children. Nevertheless, it should be noted that not all school teachers were able to perform screening: some found it difficult to understand the procedure and others lacked spare time in their busy schedules.

Conclusions

The major objective of the study was to investigate the effectiveness of smartphone-based hearing screening by school teachers among primary school children. It was found that the Hearing Test app resulted in slightly higher but significantly different thresholds than an audiometer. However, hearing thresholds obtained using the smartphone application were within normal limits. Hence, it can be concluded that it is possible for schoolteachers to utilize the Hearing Test application to identify hearing loss among primary school children. The findings suggest that hearing screening by teachers could help with the early detection of hearing loss in school children, increase the incidence of referral, and make testing more practical and affordable.

Funding

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Conference reports

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REPORT ON THE 36TH WORLD CONGRESS OF AUDIOLOGY (WCA), 19–22 SEPTEMBER 2024, PARIS, FRANCE

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The World Congress of Audiology 2024 took place from September 19 to 22 in Paris, bringing together world-class experts in hearing impairment, vestibulocochlear disorders, tinnitus, and related fields. This prestigious scientific event provided a valuable platform for knowledge exchange with renowned researchers and the presentation of the latest research and practical advances in diagnosing, treating, and managing hearing disorders. Among the attendees were thousands of specialists from around the world, including representatives from the Institute of Physiology and Pathology of Hearing (IFPS) – Prof. Piotr H. Skarzynski, MD, PhD, Prof. Artur Lorens, PhD, Dr Monika Matusiak, MD, PhD, Dr Anita Obrycka, PhD, Dr Adam Walkowiak PhD, and Emilia Czaplicka.

This year's conference program covered a wide range of topics that are becoming increasingly important in audiology. Key subjects included auditory implants, auditory objective measures, clinical genetics, single-sided deafness, and asymmetric hearing loss. Additionally, issues related to cognitive functioning in individuals with hearing loss were extensively discussed, along with strategies for preventing noise-induced hearing loss.

On the first day of the conference, during a session on Basic and Translational Research, Dr Monika Matusiak presented a lecture titled "Genetic polymorphisms of MMP9 and BDNF as biomarkers of neuroplasticity in prelingual deafness treatment by cochlear implantation." The study focused on genetic polymorphisms that may influence brain neuroplasticity in children with congenital deafness who are undergoing cochlear implantation. The results suggest that specific genetic markers could serve as predictors of auditory rehabilitation outcomes.

A highlight was the keynote session, Central Markers of Hearing Restoration, by Prof. Karen Gordon. She presented research on brain development in children with cochlear implants and unilateral deafness. Her findings demonstrated that earlier implantation reduces the risk of cortical reorganization due to auditory deprivation, thereby supporting the development of speech and language.

On the second day, Dr Walkowiak presented his research during the session on objective auditory measurements with a lecture titled "Validation of SPL chirp for ECochG measurement." His study focused on the validation of new chirp stimuli used in electrocochleography (ECochG), and showed that these stimuli could significantly improve the accuracy of diagnosing patients with hearing disorders. They could also allow better monitoring of cochlear implantation.

Dr Anita Obrycka presented her work during a session on pediatric cochlear implants. Her presentation, "Longitudinal observation of benefit after sequential bilateral cochlear implantation in prelingually deaf children", focused on the long-term benefits of sequential implantation in children with congenital deafness, emphasizing the importance of early intervention and bilateral auditory stimulation for optimal child development.

On the third day, a key event was a panel discussion on surgical considerations for auditory implants, led by Prof. Piotr Skarzynski. His lecture, "Residual hearing and inner ear malformation", addressed the challenges of cochlear implantation in patients with inner ear malformations. He highlighted the importance of preserving residual hearing during surgery and discussed techniques to minimize the risk of hearing loss.

In a session dedicated to the International Classification of Functioning, Disability, and Health (ICF), Prof. Artur Lorens presented a talk, "The protocol of outcome assessment after cochlear implantation based on the International Classification of Functioning, Disability and Health", in which he outlined a new approach to evaluating outcomes in cochlear implant patients. The ICF-based protocol offers a comprehensive assessment of patients, taking into account both auditory functions and everyday activities.

On the final day, a session chaired by Adrian Fuente focused on ototoxicity. The discussions covered the prevention and management of ototoxicity caused by various substances, including aminoglycosides and industrial chemicals. Following six presentations, there was a discussion on the role of audiologists in treating and monitoring ototoxicity.

In the Newborn Hearing Screening session, Dr Feri Zhao gave an outstanding presentation on an AI-based diagnostic tool for detecting otitis media with effusion (OME) in children, a tool which gives a sensitivity of over 90%.

Other important topics covered at the conference included the challenges of bone conduction hearing aids for children and the benefits of bimodal stimulation.

The conference also provided an opportunity to showcase the latest audiological technologies. Med-El demonstrated its new Sonnet 3 sound processor, which is waterproof, smaller, and lighter than its predecessors, significantly improving patient comfort. The processor also offers direct sound streaming, making daily life easier for cochlear implant users.

Delegates from the Institute of Physiology and Pathology of Hearing presented 16, including:

1. Effectiveness of bone conduction hearing aids in young children with congenital aural atresia and microtia.
2. Cochlear implantation in children with congenital malformations of the mastoid process.
3. A case report of riboflavin treatment and cochlear implants in a 4-year-old girl with progressive hearing loss and delayed speech development: Brown–Vialetto–Van Laere syndrome.
4. Bronchio-oto-renal syndrome: a case report.
5. Symptoms of auditory processing disorders (APD) in children with tinnitus.
6. Normative values for test of central auditory processing disorder in children aged from 6 to 12 years old.
7. Treatment of hearing loss with stapedotomy in a patient with Ehlers–Danlos syndrome.
8. The Bonebridge BCI 602 active transcutaneous bone conduction implant in children: objective and subjective benefits.
9. Stapedotomy in congenital stapes ankylosis with mobile footplate.
10. Results of surgical treatment of unilateral and bilateral otosclerosis in children.
11. Effectiveness of surgical approach of insertion ventilation tubes (tympanostomy) and adenoidectomy in comparison with non-surgical approach (watchful waiting approach) in children at the age between 1–6.
12. Multifrequency ECochG intraoperative monitoring during cochlear implantation: surgical considerations.
13. Perception of social support by adults scheduled for cochlear implantation.
14. The relationship between the electrically evoked stapedius reflex threshold and stimulus burst duration in pediatric cochlear implant users: preliminary data.
15. Binaural benefit of cochlear implant in children with single side deafness.
16. Can an unaided localization ability be a predictor of CI squelch benefit in patients with residual hearing in the implanted ear?

The event concluded with a gala event at Paris's oldest cabaret, Paradis Latin, built by Gustave Eiffel in 1889. Participants enjoyed the spectacular L'Oiseau Paradis show, featuring dancers, actors, singers, and acrobats. This event was a successful and inspiring conclusion to the conference, which showcased the latest scientific advancements in otolaryngology and audiology.

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REPORT OF THE 7TH CONGRESS OF THE CONFEDERATION OF EUROPEAN ORL-HNS, 15–19 JUNE 2024, DUBLIN, IRELAND

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The 7th Congress of the Confederation of European ORL-HNS was held in Dublin from June 15–19, 2024. The conference was attended by more than 3,400 delegates from almost all continents. This is the largest ENT meeting in Europe and was held under the patronage of the President of the VII European Congress of the ORL-HNS, John Russel, and under the scientific chairmanship of Michael Kuo. It was organised jointly with the American Academy of Otolaryngology, Head and Neck Surgery.

During the 5 days of the conference, 147 medical sessions were held across 17 rooms. The scientific program was divided into individual sessions: general, otology/otoneurology, rhinology, laryngology, phoniatry, oncology, pediatrics, salivary gland diseases, skull base and head and neck surgery, facial plastic surgery, sleep medicine, and an educational track. The sessions took the form of lectures, round tables, discussions, mentoring sessions, and instructional courses. The congress was an excellent platform for sharing experiences, mentoring education, recognising the achievements of individual European and world ENT centres, announcing otolaryngological innovations, and reporting research that will change the face of modern otolaryngology.

The team of the Institute of Physiology and Pathology of Hearing was represented by Prof. Piotr H. Skarzynski, MD, PhD, and Anna Piecuch, MD. They presented two papers: *Hearing preservation after cochlear implantation with the lateral wall electrode – outcomes in adult patients after 12 months* (by P.H. Skarzynski, A. Walkowiak, E. Bukato, A. Lorens, A. Obrycka, and H. Skarzynski) and *Early auditory development of cochlear implanted children with sensorineural hearing loss following congenital CMV infection* (by P.H. Skarzynski, A. Obrycka, A. Kolodziejak, E. Gos, A. Lorens, R. Zdanowicz, and H. Skarzynski). There was also an e-poster: *Treatment of hearing disorders in Brown–Vialetto–van Laere syndrome with cochlear implants and a case report of BVVL from Poland* (by A. Piecuch, P.H. Skarzynski, and H. Skarzynski).

Prof. Piotr H. Skarzynski presenting his lecture

The next meeting, the 8th European Congress of ORL-HNS, will take place in Gothenburg in Sweden, 25–29 April 2026 under the patronage of Congress President Ann Hermansson and should be regarded as a must.

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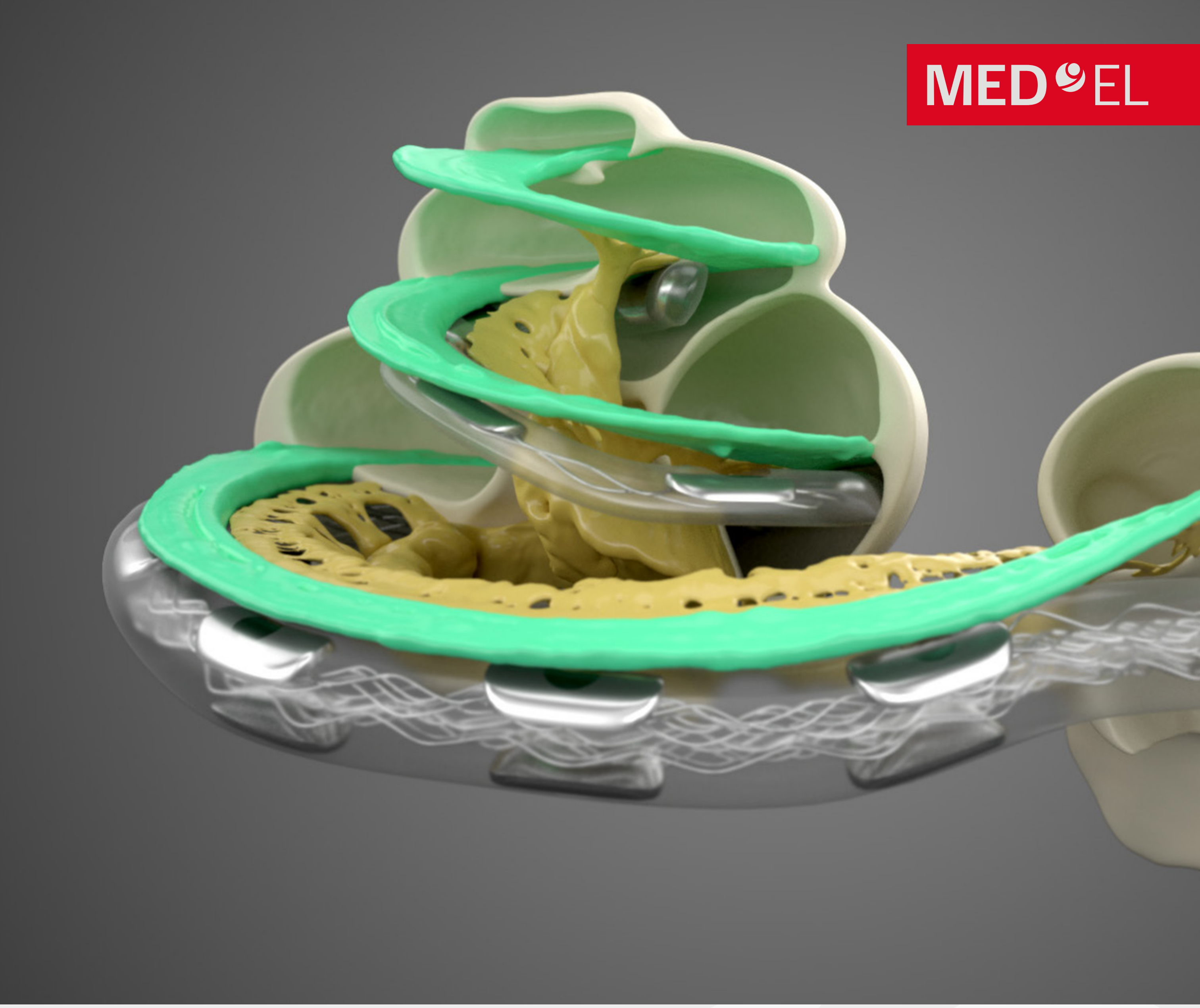
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World Hearing Center

OF THE INSTITUTE OF PHYSIOLOGY AND PATHOLOGY OF HEARING

The World Hearing Center is a modern specialized hospital providing medical care at the highest quality level in the fields of otolaryngology, audiology, phoniatrics, rehabilitation and biomedical engineering. It is superbly equipped for research and education, and includes modern conference facilities. The Center conducts a wide range of research and educational activities addressed to specialists from Poland and other countries. The Center is one of the leading medical institutions in the field of hearing disorders treatment, running, among others, one of the largest hearing implant programs in the world and performing 15,000 to 21,000 surgical procedures yearly.

The Center provides its patients with comprehensive diagnostics, conservative treatments, and surgery for the rehabilitation of:

- congenital and acquired malformations of the external, middle and inner ear,
- hearing, speech and balance disorders of different etiologies,
- disorders of the mouth cavity, throat and larynx,
- disorders of the nose and paranasal sinuses,
- sleep disorders.

World Hearing Center:

- is a global leader in terms of the number of performed otorhinolaryngological surgeries and the number of outpatient consultations (more than 200,000 consultations per year),
- is the place where unique and highly specialized medical procedures are performed, including reconstruction surgeries of congenital defects of the outer ear, treatment of profound and partial deafness with various hearing implants, phonosurgeries, endoscopic sinus surgeries under image guidance, and many others,
- employs a team of highly qualified and experienced specialists,
- has state-of-the-art medical equipment and instrumentation,
- offers comfortable conditions for hospital stays,
- uses the most modern telemedical solutions providing remote consultations via the world-first National Network of Teleaudiology.

The team of the Institute of Physiology and Pathology of Hearing and its individual employees are winners of numerous international and national awards.

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