REVIEW
OTOLOGY
DOI: 10.5152/B-ENT.2021.20195

Minimal outcome measurements in pediatric cochlear implant users: a consensus paper

Griet Mertens[1,2], Anouk Hofkens[1], Paul Van de Heyning[1,2], Vincent Van Rompaey[1,2], An Boudewyns[1,2], Maria Fernanda Di Gregorio[3], Robert H. Eikelboom[4,5,6], Roberta Marino[7,8], Anja Kurz[9], Heike Kühn[9], Wafaa Shehata-Dieler[9], Artur Lorens[10], Sasidharan Pulibalathingal[11], Ranjith Rajeswaran[12], Dayse Tavora-Vieira[7,8], Sandra R. Bellekom[4,5], Vedat Topsakal[1,2], Other HEARRING members

[1]Department of Otorhinolaryngology, Head and Neck Surgery, Antwerp University Hospital, Antwerp, Belgium
[2]Experimental Laboratory of Translational Neurosciences and Dento-Otolaryngology, University of Antwerp, Faculty of Medicine and Health Sciences, Antwerp, Belgium
[3]OTICO Hearing Center (OHC), Cordoba, Argentina
[4]Ear Sciences Centre, Faculty of Health and Medical Sciences, The University of Western Australia, Perth, Australia
[5]Ear Science Institute Australia, Subiaco, Australia
[6]Department of Speech Langauge Pathology and Audiology, University of Pretoria, South Africa
[7]Deparment of Otolaryngology, Head & Neck Surgery, Medical School, The University of Western Australia, Perth, Australia.
[8]Fiona Stanley Hospital, Perth, Australia
[9]Department of Otorhinolaryngology, Comprehensive Hearing Center, Plastic, Aesthetic and Reconstructive Head and Neck Surgery, University Hospital Würzburg, Germany
[10]Institute of Physiology and Pathology of Hearing, Kajetany, Poland
[11]ENT Super Specialty Institute and Research Center, Calicut, India
[12]Madras ENT Research Foundation (MERF), Chennai, India

Cite this article as: Mertens G, Hofkens A, Van de Heyning P, et al. Minimal outcome measurements in pediatric cochlear implant users: a consensus paper. B-ENT 2021; 17(2): 110-20.

ABSTRACT

The benefits of cochlear implantation in children with severe hearing impairments are widely known; however, there is no consensus regarding which minimal outcome measurements (MOMs) should be used to determine outcomes in this population with pediatric cochlear implant (CI). Therefore, the authors aim to propose a MOM test battery for pediatric CI recipients that can facilitate international multi-center research and collaboration. A pediatric MOM test battery was developed and agreed-upon by members of the HEARRING group across 30 expert clinics in the field of hearing implantation. The MOM test battery was chosen based on a literature search that focused on outcome measurements applied in clinical trials involving children with a hearing implant. Members of the HEARRING group were then asked to evaluate each of the pediatric MOM tests used. The final pediatric MOM test battery was defined for different chronological age categories (six weeks–18 years) at different suggested test intervals. The test battery includes objective hearing measurements, aided and unaided audiometry, speech perception tests in quiet and in noise, subjective hearing assessments, assessment of language development, and mental and motor development. This study presents a consensus on a MOM test battery for pediatric CI recipients that was agreed upon by members of the HEARRING group. This test battery should allow for international multi-center research to be able to extend and share evidence that will guide future clinical practice and research efforts in pediatric populations with CI.

Keywords: Paediatric cochlear implant recipients, cochlear implantation, minimal outcomes measurements, testing framework, standardization

Introduction

More than 50 years ago, cochlear implants (CIs) were introduced as a treatment option for individuals with severe to profound hearing loss who did not benefit (or only had a minimal benefit) from using hearing aids. Thanks to extensive research, cochlear implantation is now recognized as a safe and effective gold standard treatment for both adults and children with a severe sensorineural hearing impairment. Progressive longitudinal research continues to examine the outcomes of CI recipients with the aim of guiding future clinical practice and research efforts.

Corresponding Author: Griet Mertens, Griet.Mertens@uza.be
Received: November 30, 2020 **Accepted:** May 20, 2021
Available online at www.b-ent.be

Nonetheless, there is still a growing need for a widely used set of international quality standards on minimal outcome measurements (MOMs) to determine outcomes in CI recipients (1). Predefined MOM would allow us to monitor the auditory progress of CI recipients over time and to be able to relate on important issues such as the most ideal age for implantation and the cut-off audiological thresholds for CI indication. Moreover, the use of a predefined MOM test battery would allow for more international multi-center research studies and collaborations.

Variations in MOMs exist between different clinics and are mainly attributable to legal, reimbursement, or language differences among countries. However, many aspects of CI management can be standardized worldwide, thus improving international collaborations. Kleine Punte et al. were the first group to report a MOM test battery that mainly focused on adult CI recipients (2). The authors suggested that a MOM test battery should address a number of criteria: (1) the outcome measurements should answer the scientific criteria of simplicity, reliability, validity, and sensitivity; (2) the measurements should address dimensions that are important to clinicians and the CI recipient (symptoms, disability, and user perspective); (3) the measurements should be sensitive to meaningful changes in hearing abilities, such that they reflect the outcome of treatment or intervention; and (4) the measurements should address one of the World Health Organization (WHO) outcome domains of impairment, activities, participation, or quality of life.

As a result of new-born hearing screening, the landscape of cochlear implantation in children has shifted dramatically (3). Consequently, the number of longitudinal multi-center studies has also increased. Because hearing abilities and the age of pediatric CI recipients are crucial for choosing a MOM test battery, there exists a clinical need to develop a homogeneous MOM test battery for this population. Therefore, the HEARRING group set out to develop and agree upon a MOM test battery that could be used for pediatric CI recipients worldwide.

Methods

HEARRING group

The pediatric MOM was developed, discussed, and eventually agreed-upon by members of the HEARRING group. The HEARRING group consists of members from across 30 international expert clinics that aim to identify evidenced-based standards that can provide each potential implant recipient, regardless of age or where in the world he/she is treated, with the best possible hearing solution for their individual hearing loss. The multidisciplinary network includes surgeons, audiologists, speech and language therapists, and other skilled professionals, who collaborate as part of the HEARRING group. The global HEARRING network extends to Asia, Europe, North and South America, and Oceania.

Main Points:

- The number of longitudinal multi-center studies in pediatric cochlear implant recipients has increased with the aim of guiding future clinical practice and research efforts.
- To improve international multi-center research and collaborations, a widely used set of international quality standards on pediatric minimal outcome measurements (MOMs) is needed.
- A set of MOMs was defined for different age categories. The test battery includes objective hearing measurements, aided and unaided audiometry, speech perception tests in quiet and in noise, subjective hearing assessments, assessment of language development, and mental and motor development

Data collection

A PubMed US National Library of Medicine (http://www.ncbi.nlm.nih.gov/pubmed/) database search was performed to collect a list of MOMs that were reported in the literature. The search focused on MOMs that were applied in clinical trials that included children with cochlear implants. Experts in the field of cochlear implants were also asked to provide information on additional MOMs that are currently used. To reach a final list of MOMs, the data collected were critically discussed during the HEARRING meeting in Perth, Australia, in November 2017 using the criteria described by Bagatto et al. (4). Table 1 provides an overview of the final outcomes evaluation tools from which the final set was selected.

Results

Pediatric MOM test battery

The final pediatric MOM test battery, as chosen by the HEARRING group, is shown in *Table 2*. The final MOM test battery was divided into four chronological age categories: (1) six weeks–six months; (2) six months–two years; (3) two years–six years; and (4) six years–18 years. With 18 years being the world mean upper age for childhood, the study decided to define the pediatric population from zero to 18 years. The test intervals included assessments before implantation; three, six, and twelve months after CI activation; and yearly thereafter. The calibration of the validated test instruments should be checked routinely by appropriate experts to ensure that the equipment will produce results which meet or exceed defined criteria with a specified degree of confidence. For all measurements, the used material and test condition (i.e., best aided condition, $CI_{right\ ear}$ only, etc.) should be registered.

Although many more valuable outcome measures were considered for inclusion, the chosen set was kept as minimal as possible in order to fit in most clinical settings. Suggestions of useful additional measurements are listed in a later section (*section 8*). Interested centers should add additional outcomes to meet their specific requirements.

Case example

To provide a realistic overview of the pediatric MOM test battery, a fictitious case example will be described. This case involved a bilaterally severely hearing impaired girl who received a CI in her right ear at the age of eight months and in her left ear at the age of 16 months. Her results on the MOM test battery were registered at each suggested time interval and are shown in *Table 3*.

Table 1. Overview of the predefined selection criteria for the discussed outcome measures. The following criteria are listed: (1) Age categories for which the tests are indicated, (2) Norms: review of available standard values for pediatric population, (3) Language adoptions for use with different languages, (4) The risk for ceiling or floor effects, (5) Respondent burden referring to fact that the tool should be brief and clear enough for the caregiver to complete, (6) Discriminant validity investigating the possibility to differentiate between two subgroups who would be expected to have different scores, and (7) Sensitivity or responsiveness to measure changes in the expected direction. The outcome measures that were included in the final test battery and that were agreed upon by the HEARRING members are indicated with ✓ in the last column of the table

SELECTION CRITERIA	(1) Age categories	(2) Normative data	(3) Language adaptions	(4) Ceiling/ floor effects	(5) Respondent burden	(6) Discriminant validity	(7) Sensitivity	
OBJECTIVE MEASURES								
TEOAE	all	all	n.a.	n.a.	< 15 min.	strong	n.a.	✓
DPOAE					< 15 min.	strong	n.a.	χ
Tympanometry					< 15 min.	strong	strong	✓
Stapedius reflexes					< 15 min.	strong	n.a.	χ
ABR					± 60 min.	strong	n.a.	✓
ASSR					± 60 min.	strong	n.a.	χ
eCAP					< 15 min.	moderate	n.a.	✓
Electrical Impedance Telemetry					< 15 min.	strong	n.a.	✓
Vestibular testing					± 60 min.	strong	weak	χ
AUDIBILITY SOUNDS and SPEECH								
PTA Unaided inserts	± ≥ 9 m	NH	n.a.	n.a.	< 15 min.	strong	n.a.	✓
PTA Unaided free field	± 6 m					strong	n.a.	✓
PTA Aided monaural						strong	strong	✓
PTA Aided binaural						strong	strong	χ
Speech/sound discrimination tests (e.g. Ling 6 sounds)	± ≥ 18 m		yes	substantial skewing	< 15 min.	strong	strong	χ
SPEECH AUDIOMETRY in QUIET and in NOISE								
Unaided monaural	± ≥ 2 yr	NH	yes	n.a.	< 15 min.	strong	n.a.	χ
Unaided binaural						strong	n.a.	χ
Aided monaural						strong	strong	✓
Aided binaural						strong	strong	✓
SPEECH AUDIOMETRY in NOISE								
Fixed intensity levels	± ≥ 5 yr	NH	yes	substantial skewing	< 15 min.	moderate	moderate	χ
Adaptive intensity levels				none	< 15 min.	strong	strong	✓
SUBJECTIVE ASSESSEMENT								
LittleEARS	0 – 24 m	NH	yes	none	< 15 min.	moderate	unknown	χ
PEACH rating scale	3 m – 13 yr	NH and HI	no evidence	none	< 15 min.	unknown	unknown	χ
CAP	all	NH and HI	yes	substantial skewing	< 15 min.	moderate	moderate	✓
SIR		NH and HI	yes	substantial skewing	< 15 min.	moderate	weak	✓
LANGUAGE DEVELOPMENT								
CDI	6 m – 2 yr	NH and HI	yes	none	20 – 40 min	moderate	moderate	✓
Expressive language test	± ≥ 2 yr	depending on test	yes	none	depending on test	moderate	moderate	✓
Receptive language test			yes	none		moderate	moderate	✓
MENTAL and MOTOR DEVELOPMENT								
Bayley	6 m – 2 yr	NH	yes	none	30 – 60 min	moderate	weak	✓
SON	± ≥ 2 yr	NH and HI	yes			moderate	weak	✓

Table 2. Overview Minimal Outcome Measurements (MOM) in paediatric cochlear implant users.
[a]Otoacoustic Emissions; [b]Auditory Brainstem Response Audiometry; [c]Electrically evoked Compound Action Potential; [d]Pure Tone Average; [e]Categories of Auditory Perception Scale; [f]Speech Intelligibility Ratings; [g]MacArthur-Bates Communicative Development Inventories; [h]Reynell Developmental Language Scales; [i]Clinical Evaluation of Language Fundamentals; [j]Snijders-Oomen nonverbal intelligence tests.

PAEDIATRIC MOM CI	6 weeks - 6 months	6 months - 2 years	2 years - 6 years	6 years - 18 years
Objective measures	Pre-operatively			
	OAE[a] + Tympanometry + ABR[b]			
	Per-operatively			
	eCAP[c] + Electrical Impedance Telemetry			
	Activation, 3M, 6M, yearly			
	Electrical Impedance Telemetry			
PTA[d] Unaided	Pre-operatively, 3M, 6M, yearly			
	Inserts / free field 125 - 8000 Hz		Inserts 125 - 8000 Hz	
PTA[d] Aided	Pre-operatively, 3M, 6M, yearly			
	Warble tones Right ear$_{aided}$ only Left ear$_{aided}$ only 125 - 8000 Hz			
Speech in quiet			Pre-operatively, 3M, 6M, yearly	
			Closed set Best aided* Right ear$_{aided}$ only Left ear$_{aided}$ only 65 dB SPL	Open set Best aided* Right ear$_{aided}$ only Left ear$_{aided}$ only 65 dB SPL
Speech in noise			Pre-operatively, 3M, 6M, yearly	
			5 - 6YR	Sentences Adaptive procedure Noise fixed at 65 dB Best aided* Right ear$_{aided}$ only Left ear$_{aided}$ only
			Open set Best aided* Right ear$_{aided}$ only Left ear$_{aided}$ only	
Subjective assessment	Pre-operatively, 3M, 6M, yearly			
	LittleEARS CAP[e] SIR[f]		CAP[e] SIR[f]	
Language development		12M, yearly	Pre-operatively, 12M, yearly	
		CDI[g]	Language test	Language test
			Expressive + receptive	Expressive + receptive
			(RDLS)[h]	(CELF)[i]
Mental & motor development		Pre-operatively, 12M, yearly		
		Bayley	SON[j]	

1. Background information

The HEARRING group agreed that background information should be made available about the variables known to affect post-implantation performances (5). The child's gender and date of birth; implant information; if applicable, the presence of multiple disabilities or medical issues; mode of communication; linguistic environment (monolingual vs. bilingual); rehabilitation and school information; etiology of hearing loss; onset of hearing loss in each ear; and the type of hearing device used are indispensable for monitoring the child's progress and allow for direct comparisons within multi-center research studies. Moreover, as shown in the case example in *Table 3*, the inclusion of test dates is required to provide accurate information about the chronological age and the hearing age of the child at each of the different test intervals.

Table 3. Minimal background information for the interpretation of the minimal outcome measurements for the case example

Minimal Background Information				
Gender	**Female**	**Intervals**	**Right Ear**	**Left Ear**
Date of birth	05/06/2011	Pre-operatively	29/02/2012 C.A. 00;08,24	02/10/2012 C.A. 01;03,27
Etiology HL	Bilateral: cytomegalovirus	Implant	Synchrony pin FLEX28	Synchrony pin FLEX28
Onset HL	Bilateral: congenital			
Rehab. onset	16/04/2012 C.A. 00;10,11	Implantation	01/03/2012 C.A. 00;08,26	03/10/2012 C.A. 01;03,28
Rehab. end	27/06/2014 C.A. 03;00,22	Activation	15/03/2012 C.A. 00;09,10	20/10/2012 C.A. 01;04,15
Rehab type	Aural rehabilitation	3M post-activation	14/06/2012 C.A. 01;00,09	20/01/2013 C.A. 01;07,15
Education onset	01/09/2014 C.A. 03;02,26	6M post-activation	02/10/2012 C.A. 01;03,27	12/04/2013 C.A. 01;10,07
Education type	Mainstream school	1YR post-activation	12/04/2013 C.A. 01;10,07	12/10/2013 C.A. 02;04,07
Communication	Oral	2YR post-activation	15/04/2014 C.A. 02;10,10	30/10/2014 C.A. 03;04,25
Linguistic environment	Monolingual	3YR post-activation	02/04/2015 C.A. 03;09,27	14/10/2015 C.A. 04;04,09
Multiple disabilities	None	4YR post-activation	07/04/2016 C.A. 04;10,02	02/10/2016 C.A. 05;03,27
Medical issues	None	5YR post-activation	10/04/2017 C.A. 05;10,05	29/09/2017 C.A. 06;03,24

HL: Hearing loss, C.A.: Chronological age in the YY; MM, DD format

2. Objective hearing assessment

Tympanometry

As part of the objective assessment, tympanometry (including measures of middle ear pressure, ear canal volume, and tympanic membrane mobility) should be performed before implantation in children across all age categories in order to identify any middle ear pathologies (e.g., otitis media). Depending on the anatomy of the ear and the age of the child, a higher frequency probe tone should be used. A compliance peak within the normative values of the used equipment suggests a normal tympanic membrane mobility and middle ear pressure. Typically, the middle ear pressure is considered normal in the range of −155 to +30 daPa in children seven months of age and −165 to 45 daPa in in children 24 months of age (6). A peak outside of these limits or the absence of a compliance peak may suggest one of several pathologies. In the case example, bilateral type A tympanograms measured before implantation indicated that there is no middle ear effusion or no Eustachian tube malfunction in either ear (Figure 1).

Transient Evoked Otoacoustic Emissions (TEOAEs)

Low-intensity sounds emitted by functioning outer hair cells of the cochlea are known as otoacoustic emissions (OAEs). These emissions are caused by the energy produced by the outer hair cells in response to a brief single click stimulus that covers a broad frequency range (such as transient evoked otoacoustic emissions, TEOAEs). A probe is inserted into the ear canal containing speakers that produce sounds and a microphone to measure the resulting TEOAEs. OAE testing requires no behavioral or interactive feedback by the individual being tested. The HEARRING group decided that TEOAEs should be administered before implantation in all age categories to characterize sensitivity and functional hearing and to differentiate between the sensory and neural components of hearing loss. For the case example presented in this study, the bilateral absence of TEOAEs in the presence of A type tympanogram before cochlear implantation is suggestive of cochlear (outer hair cell) dysfunction (Figure 1). Further investigation is required to support and confirm this finding.

Auditory Brainstem Response (ABR) Audiometry.

Extensive literature searches support a strong correlation between estimated auditory brainstem (ABR) thresholds and behavioral pure-tone audiometry thresholds. As a result, ABR evaluation is widely accepted for the identification and diagnosis of hearing loss in the pediatric population (7). Therefore, ABR is essential as a preoperative objective tool to quantify the degree of hearing loss in all age categories as shown in figure 1 (8). Before implantation, no reproducible ABR responses could be found at hearing levels up to and including 90 dB nHL in the case example presented.

Evoked compound action potentials (eCAPs) of the auditory nerve

It is recommended that the evoked compound action potentials of the auditory nerve (eCAPs) be measured intraopera-

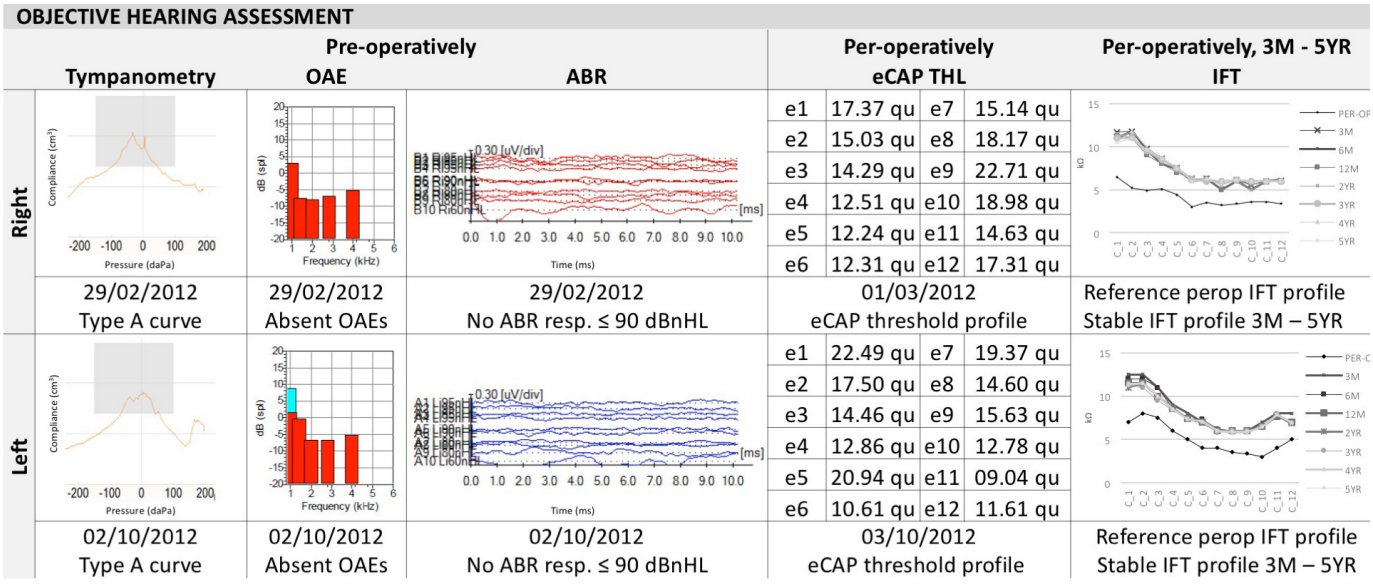

Figure 1. Results of the objective hearing assessments of the Minimal Outcome Measurements at each suggested interval for the case example. OAE: Otoacoustic emissions, M: Number of months post-activation, YR: Number of years post-activation, ABR: Auditory brainstem response, eCAP: Evoked compound action potentials of the auditory nerve, THL: Threshold level, qu: Current unit, IFT: Impedance and field telemetry

tively across all age categories (Figure 1). These measurements are frequently used to verify nerve function by stimulating one electrode contact in the cochlea and recording the resulting changes in voltage over time on another electrode contact. It is important to monitor the responses close to the round window and cochleostomy. eCAP elicited on electrodes close to the round window or cochleostomy is indicative of full insertion. The recorded eCAP measurements typically consist of a negative peak (N1) and a positive peak (P2). Although a review by Miller et al. (9) reported that the absolute values of the eCAP thresholds cannot be directly used for the prediction of the fitting parameters, the eCAP threshold profile can be used as a basis for creating fitting maps. eCAP thresholds can represent a level at which the stimulus should be audible but probably not uncomfortable. However, in addition to the eCAP profile, further fine-tuning adjustments during fitting are indispensable (10).

Electrical Impedance and Field Telemetry (IFT)

The conductivity for stimuli transmission between the surface of the electrode contact and the surrounding environment can be determined by electrical impedance measurements. Therefore, impedance telemetry of individual intracochlear electrodes can serve as an informative evaluation tool, which can provide information about efficient electrical stimulation, presence of air bubbles, extracochlear electrode positions, and open or short circuits between electrodes. Because these measurements yield important information for eCAP, eABR measurements, and audio processor programming, they should be administered to all age categories and at all test intervals. A normal IFT process was observed in the presented case example, that is, a progressive increment of IFT values during the first week after implantation, followed by a decrease and stabilization of the IFT values (Figure 1). Because no abnormal values were observed, all channels remained activated during fitting.

3. Audiometry

Although (e)ABR is a more reliable method for defining hearing thresholds in newborns and in infants up to six months of age, behavioral observation audiometry (BOA) must be added to investigate the minimal response levels in very young infants. Observing subtle unconditioned changes in behavior in response to free field sound stimuli can also be useful for parental education, particularly in terms of demonstrating the subtlety of infant hearing responses. With older children, between approximately six months to 2.5 years of age, visual reinforcement audiometry (VRA) can also be used to test their hearing thresholds. Conditioned responses to sound are recorded by reinforcing the natural tendency to turn toward a sound with a reward of an illuminated puppet or movie. From two to 2.5 years of age onwards, play audiometry can be used, whereby the child is asked to perform a simple task when they hear the sound. This may include putting a ball in a bucket or completing a puzzle. As with BOA and VRA, the volume and pitch of the sound are varied during play audiometry to determine the quietest sounds the child is able to hear. Depending on the child, ear specific information can be obtained during VRA and play audiometry.

Unaided audiometry

Due to the introduction of hearing and structure preservation into the field of cochlear implantation, the inclusion criteria for CI candidacy were expanded, resulting in greater numbers of adults and children receiving a CI. Today, individuals with some low-frequency hearing are also considered as suitable CI candidates. Therefore, it is important to evaluate, if the age and the cooperation of the child allow, the unaided residual hearing of individuals with partial deafness over time in order to offer the maximum benefits of electric acoustic stimulation (11). The limitations of the standard supra-aural headphones to measure unaided residual hearing are well described in the literature and include little exclusion of environmental background noise, the existence of cross-hearing with high-intensity stimuli presentation, the possibility of an ear canal collapse, and introduction of vibrotactile responses (12). The risk of vibrotactile responses is significantly higher in CI candidates because of the high intensity levels that are required in the low

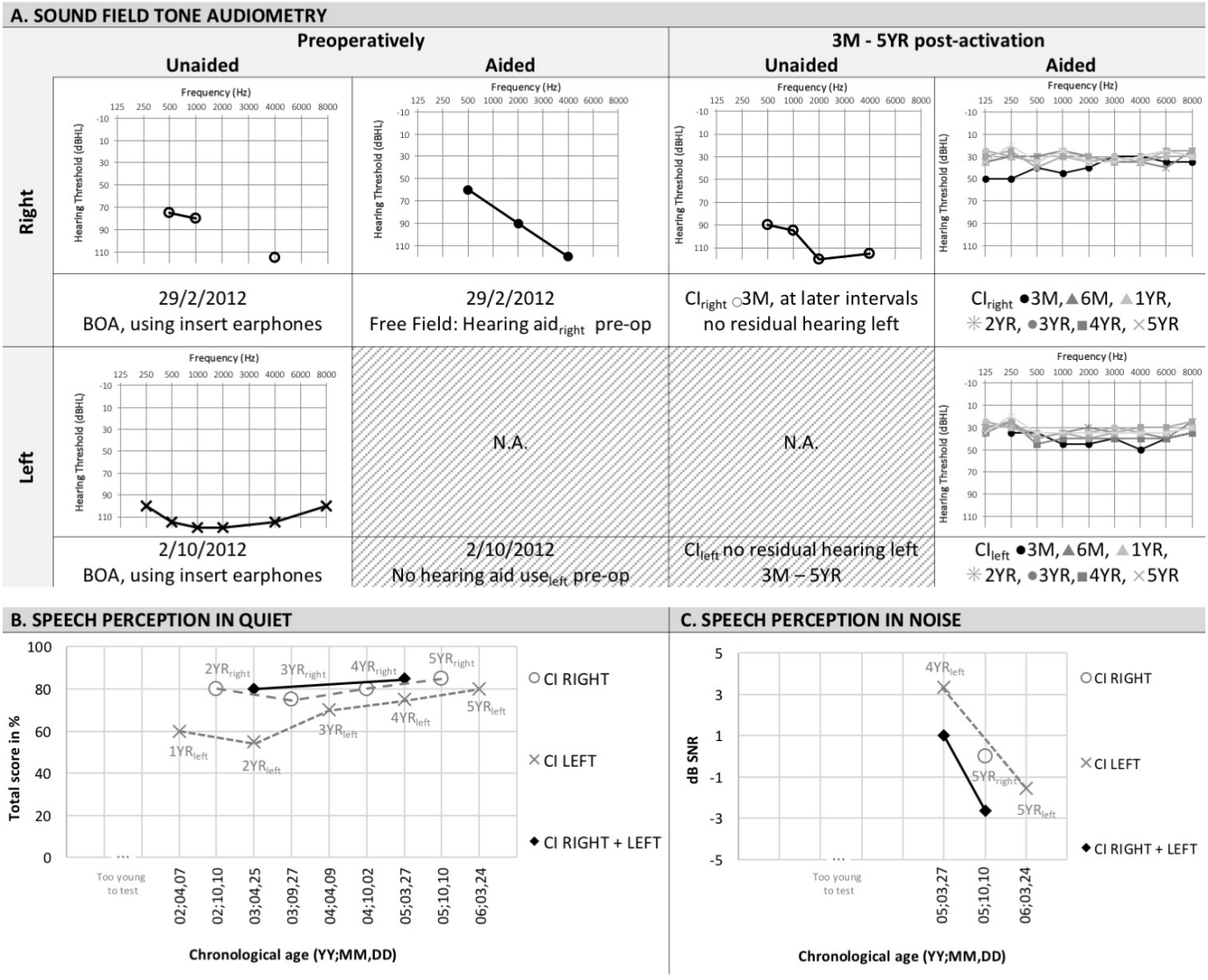

Figure 2. Audiometric results of the minimal outcome measurements at each suggested interval for the real case example. (a), Aided and unaided sound field tone audiometry. (b), Speech perception in quiet. (c), Speech perception in noise
BOA: Behavioral observation audiometry, CI: Cochlear implant

frequencies. Therefore, the HEARRING group recommends the use of insert earphones to test unaided hearing thresholds in all age categories in order to provide a solution to a number of these limitations.

Aided audiometry

Aided hearing thresholds should be measured through free field audiometry using warble tones. The loudspeaker should be placed at a distance of one meter, at head level, in front of the child. If the child wears a hearing system in both ears, then the fre

4. Speech perception

Speech audiometry is an indispensable component of the MOM test battery in children because it provides information about the understanding of speech at supra-threshold intensities (*Figure 2*). Moreover, it can be used to measure the speech, language, reading, and cognitive abilities of children. The retrieved outcomes can be used to monitor the child's progress and can support the planning and implementation of auditory rehabilitation (13). Consequently, speech perception skills must be assessed at all defined follow-up intervals using valid and reliable clinical assessment methods suitable for the pediatric CI population. The importance of speech perception testing was also discussed by Uhler et al. In 2017. They concluded that the adoption of a standardized protocol could facilitate continuity of care by constructing a Pediatric Minimum Speech Test Battery (PMSTB) (14).

In the case of bilateral hearing systems, one should start with the best aided condition, which provides the most realistic representation of the daily listening condition. Additionally, if possible, ear specific speech perception skills should also be assessed. Kosky and Boothroyd suggested that appropriate behavioral tests of speech perception performance in children should meet the following criteria: the cognitive, motoric, and attentional demands of the test should be age-appropriate; the task must be interesting and motivating; performance should be independent of vocabulary knowledge and high-

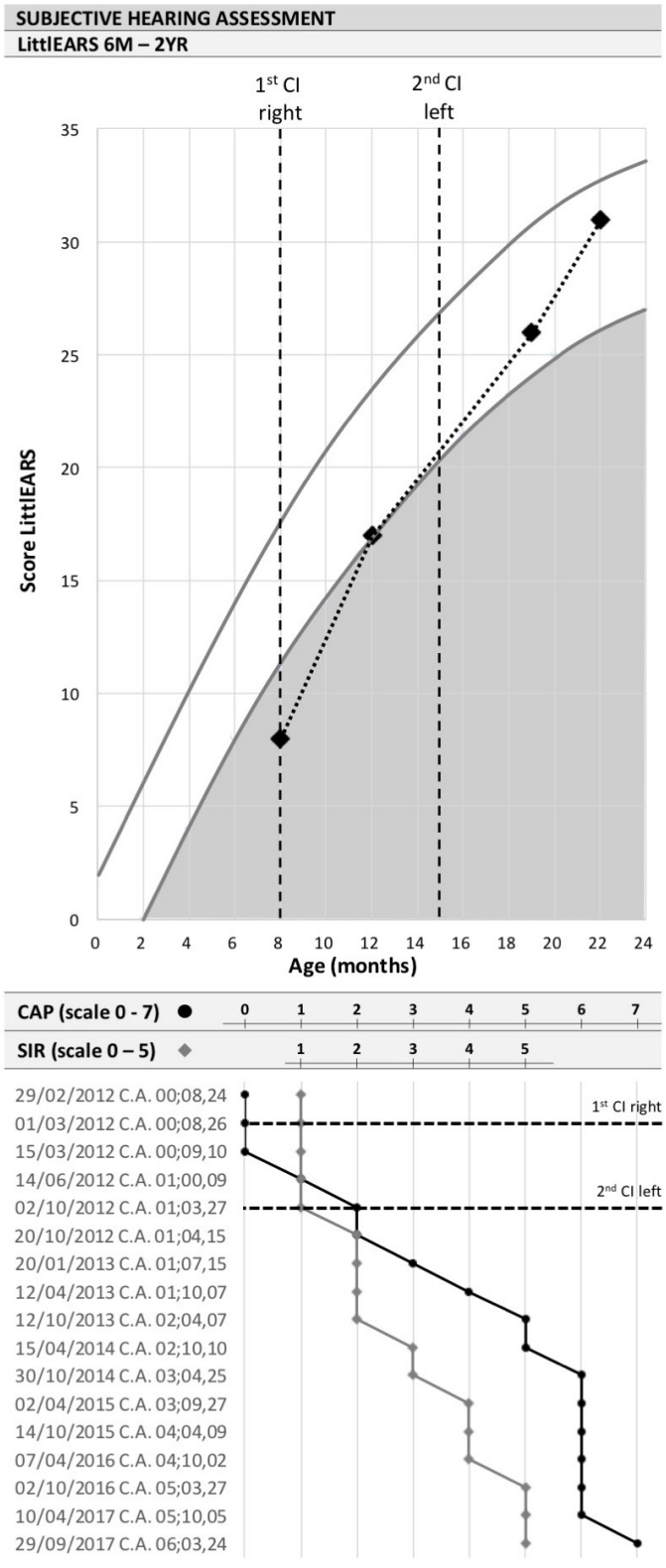

Figure 3. Results of the subjective hearing assessments of the MOM test battery at each suggested interval for the case example
M: Months post-activation, YR: Years post-activation, CI: Cochlear implant, CAP: Categories of Auditory Perception scale, SIR: Speech Intelligibility Ratings

er-level language abilities; tests should not require phonological knowledge or speech production skills; and tests should ultimately assess a person's ability to communicate in everyday situations (15). Moreover, age specific normative data should be available for the free field condition for the speech test that is used. Where possible, standardized recorded stimuli rather than live voice should be used. Live voice introduces significant variability and, for pediatric patients, may overinflate scores on speech tests (14).

Speech perception in quiet

Using age-appropriate closed-set tests, the speech perception of children between the age of two and six years should be determined at a fixed level of 65 dB SPL. Children aged six years and older should be tested with open sets at the same fixed level of 65 dB SPL, which is in accordance with the test level advised in the adult MOM test battery (2).

Speech perception in noise

Starting from the age of five years, speech perception in noise testing should be considered. Preference should be given to sentences in noise with the use of an adaptive procedure (2).

5. Subjective hearing assessment

LittlEARS

In the age categories ranging from six weeks to two years, auditory development and early speech production development of children with a hearing impairment should be assessed with the parent LittlEARS questionnaire (16). The questionnaire contains 35 "yes/no" questions and documents the receptive, semantic, and expressive behaviors that normally constitute an infant or toddler's reactions to auditory stimuli in the natural environment. In this way, the LittlEARS questionnaire should be used pre-operatively and at the post-operative test intervals to document general progress and the age appropriateness of the auditory behaviors exhibited (Figure 3)

Categories of Auditory Perception (CAP) Scale

The HEARRING group agreed that the Categories of Auditory Perception scale (CAP) should be used to measure the speech perception performance of children with CI in all age categories during all test intervals (Figure 3). The CAP measures supraliminal performance, which reflects everyday auditory performance in a more realistic way. The CAP comprises a hierarchical scale of eight performance categories arranged in order of increasing difficulty, ranging from 0 "displays no awareness of environmental sounds" to 7 "can use the telephone with a familiar talker" (17).

Speech Intelligibility Ratings (SIR)

In addition to the CAP, the speech intelligibility rating (SIR) test should be administered at all intervals in all age categories to measure the speech intelligibility of the implanted child (Figure 3). By listening to a short passage of everyday speech, speech intelligibility can be quantified using a scale between 0 and 10. The SIR consists of five performance categories ranging from ''pre-recognizable words in spoken language'' to ''connected speech is intelligible to all listeners'' (18).

6. Language development

In 1991, language development was introduced as an outcome measurement for assessing CI intervention. In the following years, language development was also used for prelingually deafened CI recipients (19). Because communication acquisition is a complex process that includes pragmatics, semantics, syntax, morphology, and phonology, not all areas can be eval-

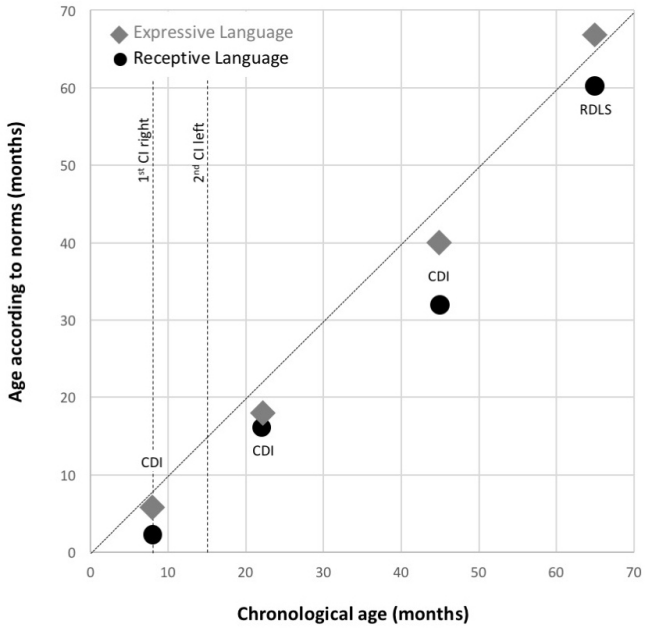

Figure 4. Results of the assessment of the language development of the case example as a part of the minimal outcome measurements at yearly follow-up intervals
CDI: MacArthur-Bates Communicative Development Inventories (age 6M – 2YR), RDLS: Reynell Developmental Language Scales (age 2YR – 6YR)

uated within a clinical set of MOMs. Therefore, the HEARRING group agreed that expressive and receptive language should at least be covered in the pediatric MOM test battery and that these areas should be evaluated before implantation and yearly thereafter using age-appropriate assessment tools (Figure 4).

MacArthur-Bates Communicative Development Inventories (CDIs)

The MacArthur-Bates Communicative Development Inventories (CDIs) can be used to assess language and communication skills between the age of six months and two years. These inventories consist of standardized parent-completed forms and a set of normative data and guidelines (20).

Reynell Developmental Language Scales (RDLS)

The Reynell Developmental Language Scales (RDLS) can be used to administer the comprehension and the production of language in children between the age of two and six years (21).

Clinical Evaluation of Language Fundamentals (CELF)

The Clinical Evaluation of Language Fundamentals (CELF) can be administered to evaluate the language abilities of school-age children (aged six years and older) over time. The CELF was designed to determine the severity of a language disorder, to identify relative strengths and weaknesses, to make recommendations regarding accommodations and interventions, and to measure the efficacy of intervention.

7. Mental and motor development

Although language development in pediatric CI recipients is the central feature of the empirical picture, mental and motor development should also be considered. There is considerable evidence that the pediatric population with a hearing impairment is vulnerable to mental and motor developmental delays. From birth onward, auditory stimulation directs visual orientation behavior. The infant's earliest responses to auditory stimuli include the visual-motor behavior of moving the eyes or head to localize sound. Consequently, it has been suggested that the lack of early auditory input could contribute to motor delays in children who are deaf or hard of hearing (22). The HEARRING group agreed that mental and motor development should be covered in the pediatric MOM test battery and that these should be evaluated before implantation and yearly thereafter using age-appropriate assessment tools.

Bayley Scales of Infant and Toddler Development

The Bayley scales are individually administered scales designed to measure the developmental functioning of infants and toddlers. Therefore, the HEARRING group recommends the use of the Bayley scales to identify possible developmental delays in the pediatric CI population between six and 24 months of age.

Snijders-Oomen nonverbal intelligence (SON) test

The Snijders-Oomen nonverbal intelligence (SON) test was developed to investigate the nonverbal intelligence of children with a hearing impairment (23). General intelligence tests were not considered because of their reliance on verbal skills. The SON test, on the other hand, covers a wide area of intelligence with nonverbal subtests related to abstract and concrete reasoning, without being dependent on the use of verbal language. Mental age norms are available for children aged two years and older.

8. Other additional measurements

Because the consensus includes only a minimal set of outcome measures, one could argue that other outcome measures not included are as least as important to meet local specific requirements. It is self-evident that interested centers should add additional outcomes to meet their specific requirements. Auditory steady-state responses (ASSRs), for example, are often added to allow frequency-specific stimulation at intensities up to 120 dB HL (instead of 95 dB HL in case of ABR testing). By adding ASSR to the clinical test battery, the clinician is able to distinguish between severe and profound hearing loss and to investigate residual hearing, which contributes to appropriate selection and fitting of hearing aids before implantation (24). Moreover, if the clinical setting allows/requires, vestibular assessment can also be considered as an essential part of the pediatric CI test battery. Sensorineural hearing loss is associated with a vestibular dysfunction in a third of the CI candidates. Additionally, cochlear implantation might have a potential impact on motor development by a (transient) vestibular deficit. It is against this background that pre- and post-operative vestibular investigation should be considered whenever possible (25).

For vestibular function testing, children 0 to 2 years of age typically receive rotary chair, cVEMP, and vHIT if a remote system is available. For children 3 to 7 years of age, vHIT, cVEMP, and oVEMP are completed, and for children 8+ years of age, vHIT, caloric testing (if vHIT is normal), cVEMP, and oVEMP are completed. Vestibular testing can be achieved with modifications tailored for the pediatric population (26).

Another outcome measure of interest is the quality of life of hearing impaired children. The HEAR-QL, for example, can serve as an excellent complement to the described MOM test battery to assess the hearing-impaired child's overall well-being (27).

Discussion

This paper describes a consensus on MOM test battery that can be used to evaluate the progress and outcomes of pediatric CI recipients. Application of a uniform test battery on MOM will also allow for international multi-center research studies to share evidence which will guide future clinical practice and research efforts in pediatric CI populations. This test battery is proposed as a part of the daily clinical practice because it only contains the minimal indispensable outcome measurements, which cover objective and subjective hearing assessments, (speech) audiometry, language, motor, and mental development. Additional testing upon individual demand is outside the scope or aim of this paper.

This MOM test battery is based on measurements that were previously applied in clinical trials that involved children with hearing implants. The test battery was developed, discussed, and eventually agreed-upon by all members of the HEARRING group. Additionally, the criteria for assessing the quality of outcome evaluation tools in rehabilitation that were previously reported in the literature were also taken into account (4, 28). The final pediatric MOM test battery was critically evaluated using the criteria described by Bagatto et al (4). All of the tests included in the test battery cover the relevant domains that were intended to be measured (i.e., hearing thresholds, speech understanding, receptive and expressive language, etc.).

The HEARRING group recommends that the calibration of the used materials should be routinely checked by appropriate experts and that age appropriate normative data should be available for each of the tests used. Moreover, the tests should be able to capture the true breadth and detail of the differences that exist within the heterogeneous pediatric population with a hearing impairment. Measurement tools with existing floor and ceiling effects were avoided insofar as possible, with ceiling effects only reported with the SIR.

The MOM test battery did not show any evidence of bias when used with children with a hearing impairment. Additionally, the results obtained were not affected by differences in culture or social circumstances. The criterion "respondent burden" was also met in the final MOM test battery with minimal patient or parental distress or burden associated with participation in the test battery. Because the test battery only contains the minimal indispensable measurements that are acceptable to both the respondent and the administrator in terms of duration and content, it can be implemented into clinical practice. Another advantage with the MOM test battery is that some of the tests can be delivered electronically or on paper and in different languages, such as the LittlEARS questionnaire.

We know from previous evidence that the included outcome measures are reliable**.** They have been shown to provide consistent results across time and testers, indicating good clinical value. Outcome measurements that were used in previous studies investigating two subgroups of the population (e.g., children with normal hearing vs. children with a hearing impairment) were chosen to be part of the MOM test battery. Therefore, the criterion for "Discriminant validity" was also met.

In conclusion, the information presented within this study proposes a basic set of MOMs that can be used for monitoring and standardizing clinical practice. Additionally, the MOM test battery can be used as a guideline for data collection and the establishment of a registry.

Peer-review: Externally peer-reviewed.

Author Contributions: Supervision – G.M., V.T.; Design – G.M., P.V., V.T.; Resources –N/A.; Materials – G.M., A.H., P.V.D.H., V.V.R., A.B., M.F.D.G., R.H.E., R.M., A.K., H.K., W.S.D., A.L., S.P., R.R., D.T.V., S.R.B., V.T., Other HEARRING members.; Data Collection and/or Processing – G.M., A.H., P.V.D.H., V.V.R., A.B., M.F.D.G., R.H.E., R.M., A.K., H.K., W.S.D., A.L., S.P., R.R., D.T.V., S.R.B., V.T., Other HEARRING members.; Analysis and/or Interpretation – G.M.; Literature Search – G.M.; Writing Manuscript – G.M.; Critical Review – G.M., A.H., P.V.D.H., V.V.R., A.B., M.F.D.G., R.H.E., R.M., A.K., H.K., W.S.D., A.L., S.P., R.R., D.T.V., S.R.B., V.T., Other HEARRING members.

Conflict of Interest: The authors have no conflict of interest to declare.

Financial Disclosure: The authors declared that this study has received no financial support.

Other HEARRING members: Rubens de Brito, Julia Speranza Zabeu, Li Yongxin, Bo Liu, Marco Caversaccio, Wilhelm Wimmer, Stefan Dazert, Stefan Vollkenstein, Christopher H. Raine, Jane Martin, Manikoth Manoj, Sasidharan Pulibalathingal, Kevin Brown, Brendan O'Connell, Meg Dillon, Mohan Kameswaran, Mario Zernotti, Timo Stöver, Uwe Baumann, Joachim Schmutzhard, Patrick Zorowka, Kurt Stephan, Hinrich Staecker, Lorne Parnes, Sumit Agrawal, Kim Zimmerman, Javier Gavilán, Luis Lassaletta, Miryam Calvi, Iain Bruce, Martin O'Driscoll, Shin-Ichi Usami, Hideaki Moteki, Shin-ya Nishio, Marcus Atlas, Peter Friedland, Aanand Acharya, Abdulrahman Hagr, Medhat Yousef, Vlad Kuzovkov, Serafima Sugarova, Henrik Smeds, Eva Karltorp, Gunnar Eskilsson, Wolfgang Gstöttner, Wolf-Dieter Baumgartner, Alexandra Jappel, Henryk Skarzynski, Piotr Skarzynski, Rudolf Hagen, Kristen Rak, Joachim Müller, Robert Mlynski, Gunesh Rajan, Georg M. Sprinzl, Benoit Godey

References

1. Bruijnzeel H, Ziylan F, Stegeman I, Topsakal V, Grolman W. A Systematic review to define the speech and language benefit of early (<12 Months) pediatric cochlear implantation. Audiol Neurootol 2016; 21: 113-26. [CrossRef]
2. Kleine Punte A, Van de Heyning P. Quality standards for minimal outcome measurements in adults and children. Cochlear Implants Int 2013; 14 Suppl 2: S39-42. [CrossRef]
3. Korver AM, Smith RJ, Van Camp G, et al. Congenital hearing loss. Nat Rev Dis Primers 2017; 3: 16094. [CrossRef]
4. Bagatto MP, Moodie ST, Seewald RC, Bartlett DJ, Scollie SD. A critical review of audiological outcome measures for infants and children. Trends Amplif 2011; 15: 23-33. [CrossRef]
5. Cosetti MK, Waltzman SB. Outcomes in cochlear implantation: variables affecting performance in adults and children. Otolaryngol Clin North Am 2012; 45: 155-71. [CrossRef]
6. Palmu A, Puhakka H, Huhtala H, Takala AK, Kilpi T. Normative values for tympanometry in 7- and 24-month-old children. Audiology 2001; 40: 178-84. [CrossRef]
7. Stapells DR. Threshold estimation by the tone-evoked auditory brainstem response: a literature meta-analysis. J Speech Lang Pathol Audiol 2000; 24: 74-83.
8. Hang AX, Roush PA, Teagle HF, et al. Is "no response" on diagnostic auditory brainstem response testing an indication for cochlear implantation in children? Ear Hear 2015; 36: 8-13. [CrossRef]

9. Miller CA, Brown CJ, Abbas PJ, Chi SL. The clinical application of potentials evoked from the peripheral auditory system. Hear Res 2008; 242: 184-97. [CrossRef]
10. Willeboer C, Smoorenburg GF. Comparing cochlear implant users' speech performance with processor fittings based on conventionally determined T and C levels or on compound action potential thresholds and live-voice speech in a prospective balanced crossover study. Ear Hear 2006; 27: 789-98. [CrossRef]
11. von Ilberg C, Kiefer J, Tillein J, et al. Electric-acoustic stimulation of the auditory system. New technology for severe hearing loss. ORL J Otorhinolaryngol Relat Spec 1999; 61: 334-40. [CrossRef]
12. Andrew Stuart RS, Tompkins C, Vandenhoff S. Test-Retest Variability in Audiometric Threshold with Supraaural and Insert Earphones among Children and Adults. Audiology 2009; 30: 82-90. [CrossRef]
13. Mendel LL. Current considerations in pediatric speech audiometry. Int J Audiol 2008; 47: 546-53. [CrossRef]
14. Uhler K, Warner-Czyz A, Gifford R, Working Group P. Pediatric minimum speech test battery. J Am Acad Audiol 2017; 28: 232-47. [CrossRef]
15. Kosky C, Boothroyd A. Validation of an on-line implementation of the Imitative test of Speech Pattern Contrast perception (IMSPAC). J Am Acad Audiol 2003; 14: 72-83. [CrossRef]
16. Coninx F, Weichbold V, Tsiakpini L, et al. Validation of the LittlEARS((R)) Auditory Questionnaire in children with normal hearing. Int J Pediatr Otorhinolaryngol 2009; 73: 1761-8. [CrossRef]
17. Archbold S, Lutman ME, Marshall DH. Categories of auditory performance. Ann Otol Rhinol Laryngol Suppl 1995; 166: 312-4.
18. McDaniel DM, Cox RM. Evaluation of the speech intelligibility rating (SIR) test for hearing aid comparisons. J Speech Hear Res 1992; 35: 686-93. [CrossRef]
19. Ruben RJ. Language screening as a factor in the management of the pediatric otolaryngic patient: Effectiveness and efficiency. Arch Otolaryngol Head Neck Surg 1991; 117: 1021-5. [CrossRef]
20. Fenson L, Dale PS, Reznick JS, et al. MacArthur-Bates communicative development inventories. Available at: https://mb-cdi.stanford.edu/index.html on July 2018. 1993.
21. Reynell J. A developmental approach to language disorders. Br J Disord Commun 1969; 4: 33-40. [CrossRef]
22. Gheysen F, Loots G, Van Waelvelde H. Motor development of deaf children with and without cochlear implants. J Deaf Stud Deaf Educ 2008; 13: 215-24. [CrossRef]
23. Snijders J. [Subjects of normal hearing and deaf mutes in the Snijders-Oomen test series]. Nederlands tijdschrift voor de psychologie en haar grensgebieden 1955; 10: 472-84.
24. Beck RM, Grasel SS, Ramos HF, et al. Are auditory steady-state responses a good tool prior to pediatric cochlear implantation? Int J Pediatr Otorhinolaryngol 2015; 79: 1257-62. [CrossRef]
25. Thierry B, Blanchard M, Leboulanger N, et al. Cochlear implantation and vestibular function in children. Int J Pediatr Otorhinolaryngol 2015; 79: 101-4. [CrossRef]
26. Janky KL, Rodriguez AI. Quantitative vestibular function testing in the pediatric population. Semin Hear 2018; 39: 257-74. [CrossRef]
27. Umansky AM, Jeffe DB, Lieu JE. The HEAR-QL: quality of life questionnaire for children with hearing loss. J Am Acad Audiol 2011; 22: 644-53. [CrossRef]
28. Andresen EM. Criteria for assessing the tools of disability outcomes research. Arch Phys Med Rehabil 2000; 81: S15-20. [CrossRef]