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Functional Hearing Quality in Prelingually Deaf School-Age Children and Adolescents with Cochlear Implants

William G. Kronenberger1,2, **Hannah Bozell**2, **Shirley C. Henning**2, **Caitlin J. Montgomery**2, **Allison M. Ditmars**2, **David B. Pisoni**2,3

¹Department of Psychiatry, Indiana University School of Medicine, Indianapolis, Indiana, USA

²Department of Otolaryngology – Head and Neck Surgery, Indiana University School of Medicine, Indianapolis, Indiana, USA

³Department of Psychological and Brain Sciences, Indiana University, Bloomington, Indiana, USA

Abstract

Objective: This study investigated differences in functional hearing quality between youth with cochlear implants (CIs) and normal hearing (NH) peers, as well as associations between functional hearing quality and audiological measures, speech perception, language, and executive functioning.

Design: Youth with CIs and NH peers completed measures of audiological functioning, speech perception, language, and executive functioning. Parents completed the Quality of Hearing Scale (QHS), a questionnaire measure of functional hearing quality.

Study Sample: Participants were 43 prelingually-deaf, early-implanted, long-term CI users and 43 NH peers aged 7-17 years.

Results: Compared to NH peers, youth with CIs showed poorer functional hearing quality on the QHS Speech, Localization, and Sounds subscales and more hearing effort on the QHS Effort subscale. QHS scores did not correlate significantly with audiological/hearing history measures but were significantly correlated with most speech perception, language, and executive functioning scores in the CI sample. In the NH sample, QHS scores were uncorrelated with speech perception and language and were inconsistently correlated with executive functioning.

Conclusions: The QHS is a valid measure of functional hearing quality that is distinct from office-based audiometric or hearing history measures. Functional hearing outcomes are associated with speech-language and executive functioning outcomes in CI users.

Keywords

deafness; cochlear implant; hearing; speech perception; memory; listening effort

Address correspondence to William Kronenberger, Ph.D., DeVault Otologic Research Laboratory, Department of Otolaryngology – Head and Neck Surgery, 699 Riley Hospital Drive, Riley Research Building 132, Indianapolis, IN 46202. Phone: (317) 274-4915. Fax: (317) 274-4949. wkronenb@iupui.edu.

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Cochlear implants (CIs) restore some components of hearing to deaf children, allowing for access to sound and the development of spoken language skills (Geers, Brenner, & Tobey, 2011; Niparko et al., 2010). The efficacy of CIs for outcomes ranging from hearing/ audiological measures (Wolfe & Schafer, 2017) to speech perception (Davidson, Geers, Blamey, Tobey, & Brenner, 2011) and higher-order language skills (Geers & Sedey, 2011; Kronenberger & Pisoni, 2019) is well-established, although some spoken language skills, such as speech perception in noise, are particularly challenging for CI users (Davidson et al., 2011). Furthermore, research has demonstrated that as many as 30-50% of children with CIs are at risk for delays in some domains of neurocognitive development, particularly executive functioning (EF; brain-based self-regulatory processes necessary to direct and deploy mental resources in order to maintain focus and achieve goals in the face of distraction), including working memory (holding information in immediate memory during additional concurrent cognitive demands), inhibition-concentration (resisting impulses and delaying behavior to consider consequences), and controlled fluency-speed (remaining focused on an activity in order to complete it rapidly and efficiently) (Kronenberger & Pisoni, 2018). Over all, considerable variability in auditory, speech perception, language, and neurocognitive outcomes is found within the population of children with CIs, and explaining that variability is one of the most important clinical and scientific issues in the cochlear implantation field (Pisoni, Kronenberger, Harris, & Moberly, 2018).

Access to auditory sensory experience and information after cochlear implantation is a prerequisite for the development of robust spoken language skills. As a result, measures of audiological functioning and hearing history have been some of the earliest and most investigated predictors of speech perception and language outcomes. Greater exposure to sound prior to implantation (reflected by measures such as pre-implantation unaided pure tone average (PTA), age at onset of deafness, and duration of deafness) is often (although not universally) associated with better speech perception and language outcomes (Geers, Brenner, & Davidson, 2003; Geers et al., 2011; Geers, Nicholas, & Sedey, 2003; Geers & Sedey, 2011; Ruffin, Kronenberger, Colson, Henning, & Pisoni, 2013). Furthermore, greater quality and quantity of aided auditory experience (measured by aided PTA, age at implantation, and years of CI use) following implantation is related to better speech and language outcomes (Geers, Brenner, et al., 2003; Geers et al., 2011; Geers, Nicholas, et al., 2003; Geers & Sedey, 2011; Niparko et al., 2010; Ruffin et al., 2013). However, these commonly used conventional measures of audiological functioning and hearing history before and after implantation do not fully explain the enormous variability and individual differences routinely observed in spoken language outcomes following cochlear implantation (Pisoni et al., 2018). Furthermore, measures of audiological functioning and hearing history typically are minimally related (if at all) to neurocognitive and executive functioning outcomes in children and adolescents with CIs (Kronenberger, Pisoni, Henning, & Colson, 2013). The failure of audiological and hearing history variables to sufficiently predict speech, language, and neurocognitive outcomes following cochlear implantation has led to a robust search for additional predictors of outcome (Kronenberger & Pisoni, 2020).

Almost all of the research investigating audiological predictors of spoken language and neurocognitive outcomes following implantation has focused on audiometric (typically preimplant and aided post-implant PTA), device (e.g., number of active electrodes, type of

processor and strategy), and hearing history (age of onset of deafness, age at implantation, duration of deafness) variables. However, an additional critical domain of auditory outcome is functional hearing quality (also referred to as functional auditory performance or functional auditory measurement). Functional hearing quality refers to the quality of speech and sound perception in the variety and complexity of everyday environments, and measures of functional hearing quality are recommended as a part of routine assessments after cochlear implantation (Wolfe & Schafer, 2017).

Functional hearing quality is typically assessed by asking a rater how well a person hears in different situations, sounds, and contexts, such as speech in a quiet room or sounds when outside, using a questionnaire or interview format (Galvin & Noble, 2013; Kopun & Stelmachowicz, 1998). For adults, ratings are made based on self-report, whereas for children, ratings may be made by parents, teachers, or other caregivers, based on observations of the child's functioning and behavior. Item responses may be grouped into subscales indicating subdomains of functional hearing, such as hearing in quiet situations, in noise, for speech, for environmental sounds, and for localization of sounds (Bagatto et al., 2019; Ching & Hill, 2007; Tharpe & Flynn, 2016).

Several interviews and questionnaires have been developed to measure functional hearing quality in children (see (Bagatto et al., 2019) and (Ching & Hill, 2007) for lists and descriptions). These measures differ along several parameters: age range, questionnaire vs. interview format, target population (e.g., hearing aid users, cochlear implant users, etc.), respondent/rater (e.g., parent, teacher, child), and content. For measures that cover the broad childhood age range from preschool through adolescence, use of a parent or teacher as the rater (in lieu of self-report) is essential because of limited self-awareness, reading ability, comprehension, and insight in children prior to early adolescence.

Despite the availability of several measures of functional hearing quality in children, there has been relatively little peer-reviewed research investigating the psychometrics of those measures or their association with hearing, speech, language, and neurocognitive outcomes. In one study, parents of children with varying degrees of hearing loss completed the Auditory Behavior in Everyday Life (ABEL) questionnaire, which provides 3 scales measuring Aural-oral (speaks/sings/responds to social stimuli), Auditory awareness (responds to speech or sounds), and Social/Conversational skills (social communication) (Purdy, Farrington, Moran, Chard, & Hodgson, 2002). Results from a small sample of CI users (N=7) showed improvement in ABEL scores over time after implantation (Purdy et al., 2002). In another study, parents of 7 children with CIs and integrated electric-acoustic sound processors completed the children's version of the Speech Spatial Qualities (SSQ-C (Galvin & Noble, 2013)) and the Children's Home Inventory for Listening Difficulties (CHILD, which yields a score for listening skills at home in 3-12 year old children) (Wolfe et al., 2017). The SSQ-C is an adaptation of the well-established adult SSQ for child-, parent-, or teacher-report, which is administered as an interview and yields data for the same domains as the adult SSQ. Results demonstrated improvements in subjective hearing with use of electric-acoustic sound processors.

In the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study, the Parents' Evaluation of Aural/Oral Performance of Children (PEACH (Ching & Hill, 2007)) was used to assess parent-rated functional hearing (in quiet or in noisy settings) of children aged preschool-7 years with hearing aids or CIs (Ching, Dillon, Leigh, & Cupples, 2018; Ching et al., 2013). Results demonstrated that PEACH scores were associated with language scores and that higher PEACH scores were associated with better psychosocial outcomes (Ching et al., 2018; Ching et al., 2013). Studies using the Abbreviated Profile of Hearing Aid Performance (APHAP) (Kopun & Stelmachowicz, 1998) have shown modest agreement between parent- and child-report questionnaires, with correlations in the 0.13 to 0.47 range. Parent-report APHAP subscales have been inconsistently related to audiometric and speech perception measures (Hillock-Dunn, Taylor, Buss, & Leibold, 2015).

In summary, existing research using parent-report measures of functional hearing quality has demonstrated that functional hearing quality is a multifactorial construct consisting of domains including quiet/noise and speech/sounds, which can be reliably and validly assessed based on parent-report. Functional hearing quality ratings are related to audiometric measures and diagnosis of hearing loss, although functional hearing quality is not fully explained by audiometric values. Furthermore, functional hearing quality scores can be used to validly assess outcomes after interventions such as cochlear implantation. In addition to parent-report questionnaires of functional hearing quality, other functional hearing quality measures exist for more restricted age ranges, for teacher-report of functional hearing quality, and for use as interviews (Bagatto et al., 2019; Ching & Hill, 2007; Tharpe & Flynn, 2016). Additionally, adult self-report measures have been used to test and document functional hearing quality in adults with hearing aids and CIs, both as a separate construct and as a component of quality of life (McRackan, Hand, Velozo, & Dubno, 2019).

Despite the number and types of functional hearing quality measures, there still remains a dearth of knowledge about the functional hearing outcomes of prelingually-deaf school-age children and adolescents with CIs. Prior research on functional hearing outcomes of children with CIs has used small samples or has combined CI users and hearing aid users. Additionally, there is almost no literature investigating the association of functional hearing outcomes with speech-language and neurocognitive outcomes in CI users, despite the critical underlying importance of hearing for those domains of functioning.

The purpose of the present study was to investigate the association between functional hearing quality and measures of audiological, hearing history, speech perception, language, and executive functioning in early-implanted school-age children and adolescents with CIs. In order to accomplish this goal, we sought to identify a measure with the following characteristics:

- **1.** Questionnaire-based, brief, and easy to complete.
- **2.** Suitable for parent-report.
- **3.** Appropriate for use with samples of CI users as well as NH peers as a comparison.

- **4.** Appropriate for school ages (7-17 years), but for future work also applicable to preschool and young adult populations.
- **5.** Multifactorial, with subscales reflecting not only hearing quality for sounds, but also speech and hearing effort.

No existing measure of functional hearing quality encompassed all of these characteristics. The SSQ and PEACH, for example, are well-validated, but are designed to be administered in interview format. Furthermore, the PEACH is designed for the preadolescent age range (preschool-7 years). The ABEL has been used with only a small number of CI users and has limited psychometric data, while the APHAP was designed primarily for children with hearing aids. Other measures of functional hearing quality (Bagatto et al., 2019; Ching & Hill, 2007; Tharpe & Flynn, 2016) have not been subjected to extensive psychometric testing and/or have seen little use or validation in samples of CI users. Finally, no existing parent-report measure of functional hearing quality for children had an effort subscale. The inclusion of an effort subscale was important for the present analyses because of the importance of understanding relations between listening effort, speech, language, and neurocognitive functioning in children with hearing loss (Pichora-Fuller et al., 2016; Rönnberg et al., 2013). Because no existing measure of functional hearing quality met all of these criteria, a secondary goal of this study was to develop and validate a new parent-report, questionnaire-based measure of hearing quality for use with youth and young adults with CIs.

Thus, the primary hypotheses of the current study were as follows:

- **1.** School-age children and adolescents with CIs will demonstrate poorer parentreported functional hearing quality and will use more effort during functional hearing tasks compared to same-aged NH peers.
- **2.** Functional hearing quality will show modest but statistically significant associations with audiological and hearing history measures in children with CIs.
- **3.** Functional hearing quality will correlate significantly with measures of speech perception and language in children with CIs, and these correlations will be stronger than those for NH peers, reflecting the greater importance of hearing quality for speech-language development in CI users.
- **4.** Functional hearing quality will be associated with measures of executive functioning in CI users, consistent with theories linking hearing, language, and executive functioning in CI users (Kronenberger & Pisoni, 2020).

Method

Participants

Participants for the present study were 43 children with CIs and 43 NH peers. One parent for each child also completed questionnaires of functional hearing quality and executive functioning (36 mothers, 5 fathers, and 2 other female caretakers for the CI sample; 40 mothers and 3 fathers for the NH sample). Inclusion criteria for the sample of CI users were: (1) hearing loss prior to age 3 years; (2) cochlear implantation at age 3 years, 11 months or

younger; (3) enrolled or living in an environment that encouraged the development and use of spoken language skills; and (4) use of CI for at least 7 years. Inclusion criteria for the sample of NH peers were: (1) normal hearing and language by parent-report; and (2) hearing screening within normal range (each ear individually tested at frequencies of 500, 1000, 2000, and 4000 Hz at 20 dB HL using Telephonics TDH-50P headphones in an Acoustic Systems RE243 soundbooth). Inclusion criteria for both samples were: (1) absence of developmental, cognitive, or neurological diagnoses; (2) home environment in which spoken English is the primary language; (3) nonverbal IQ greater than 2 standard deviations below the normative mean (defined by Leiter-3 Classification and Analogies subtest scaled score of 4 or greater); and (4) age 7-17 years.

Procedures

Study procedures were approved by the local Institutional Review Board. Written consent and assent were obtained prior to administration of study procedures. Testing was completed in a single session lasting approximately 3 hours, during which children were individually tested by ASHA-certified speech-language pathologists, while one parent completed questionnaires in a separate room. All tests and directions in the present study were administered in the same order using standard instructions across the full age range of the study (with breaks as needed), consisting of live-voice presentation in auditory-verbal format without the use of any sign language and with the examiner's face in view, with the exception of speech recognition tests administered via audiorecording. Audiorecorded items were presented in a quiet setting at 65 dB sound pressure level (SPL) using a high quality loudspeaker located approximately 3 feet from the particpant. On a 1 (not at all) to 7 (totally) examiner rating scale of test validity, all participants were rated 5 or higher, and 93% of participants in each (CI and NH) subsample received a validity rating of 6 or 7.

Measures

Audiological and Hearing History (CI sample only).—Audiological and hearing history variables obtained from the medical record and/or parent interview for the CI sample (this data was not available for some subjects from medical records review) consisted of age of onset of deafness (N=41), duration of deafness (N=41), age of implantation (N=41), duration of CI use (N=41), unaided pre-implant pure tone average (PTA) in the best ear for frequencies 500, 1000, and 2000 Hz in dB HL (N=39), and most recent aided post-implant PTA in the best ear (N=29) (Table 1).

Quality of Hearing Scale (QHS).—The QHS is a 21-item questionnaire asking parents to report on their child's hearing experiences in four domains: Speech, Localization, Sounds, and Effort (see Table 2 for item and subscale content; a copy of the QHS is available from the authors). Parents rate on a 0 (not at all) to 10 (a lot; completely) scale (with anchors of "a little" at a "2" rating, "some" at a "4" rating, "pretty much" at a "6" rating, and "very much" at an "8" rating) how well their child hears or how much effort he/she uses for hearing in a variety of settings and situations. Items for the QHS were selected based on literature review and consultation with clinicians who work with children who have cochlear implants. Raw QHS subscale scores are averages of constituent items on subscales (based on the four domains above), and a QHS Total score is obtained by averaging the four subscale scores

greater listening effort).

Speech Perception/Sentence Recognition.—Four measures were used to assess different domains of speech perception and sentence recognition: the Children's Test of Nonword Repetition (CNREP; nonword repetition) (Gathercole & Baddeley, 1996), the Lexical Neighborhood Test (LNT; spoken word recognition) (Kirk, Pisoni, & Osberger, 1995), and the Hearing in Noise Test for Children (HINT-C; sentence recognition) (Nilsson, Soli, & Gelnett, 1996) administered in quiet and in noise. For the CNREP, participants repeated spoken nonwords presented via audio recording; percentage of whole nonwords reproduced correctly was the dependent measure used in the present data analyses. The LNT is an open-set word recognition test of monosyllabic words presented via audio recording. Subjects are presented with a lexically easy or lexically hard word and must repeat the word as they heard it. Lexically easy words are words that are phonologically distinctive (share few phonemes with other words in the lexicon), whereas lexically hard words are more phonologically confusable with other words (Kirk et al., 1995). For the present study, LNT score was percentage of total words correct. The HINT-C is an open-set sentence recognition test consisting of meaningful spoken sentences presented via audio recording in quiet and in speech-shaped noise (+5 dB signal-to-noise ratio). Separate scores for quiet and noise are obtained from percentage of keywords correctly repeated by subjects. For the NH sample, only the CNREP was obtained because NH children and adolescents routinely score at the ceiling on the LNT and HINT-C.

Language.—Vocabulary knowledge and understanding/memory of verbal directions were assessed with the Peabody Picture Vocabulary Test, Fourth Edition (PPVT-4) (Dunn & Dunn, 2007) and the Clinical Evaluation of Language Fundamentals, Fifth Edition (CELF-5) (Semel, Wiig, & Secord, 2013) Following Directions subtest, respectively. For the PPVT-4, participants are asked to identify which of four stimulus pictures best represents the meaning of a single word spoken by the examiner; age norm-based standard score was used in the present analyses. For the CELF-5 Following Directions subtest, the examiner provides a set of spoken directions that the participant follows; age norm-based scaled scores were used.

Nonverbal Intelligence.—Nonverbal intelligence was measured with the Classification and Analogies subtest of the Leiter International Performance Scale, Third Edition (Leiter-3) (Roid, Miller, Pomplun, & Koch, 2013), which requires subjects to place items in a tray based on conceptual classification or analogic relationships between designs. Age-normbased scaled scores were used to assess nonverbal/fluid intelligence.

Executive Functioning.—Verbal working memory was assessed using two individuallyadministered measures: (1) a Computerized Visual Digit Span (VDS) test and (2) the Letter-Number Sequencing (LNS) subtest of the Wechsler Intelligence Scale for Children, Fifth Edition (WISC-V) (Wechsler, 2014). For the VDS, a sequence of digits (1-9) is presented randomly on a computer screen at 1 second intervals, and the subject then is asked to reproduce the sequence in either forward (VDS Forward) or backward (VDS Backward) order by touching the numerals on a 3×3 response grid presented on a computer display

monitor at the end of the sequence. The number of digits in the sequence begins with 2 and increases by 1 digit after every 2 trials until 2 trials are failed at the same sequence length. Separate raw scores are obtained for VDS Forward and VDS Backward based on the number of sequences reproduced correctly. Digit span tests in general, and the VDS in particular, have been extensively validated as measures of verbal working memory capacity in children and adolescents with CIs (AuBuchon, Pisoni, & Kronenberger, 2015; Kronenberger & Pisoni, 2018). The LNS subtest is a widely used and validated measure of auditory-verbal working memory (Wechsler, 2014). For the LNS, the examiner presents (in spoken format) a series of random letters and numerals (starting with a sequence length of 2 and increasing up to a sequence length of 8); for each letter-number series, the subject is asked to repeat the numbers first in ascending order, followed by the letters in alphabetical order. Sequences of the same length are presented in groups of 3, and the test is discontinued when the subject fails all 3 items within the same group; raw scores are the number of correctly reproduced sequences.

Norm-based scaled scores from two nonverbal Leiter-R executive functioning subtests were used to assess controlled fluency-speed and inhibition-concentration (Roid et al., 2013). The Attention Sustained subtest is a cancellation subtest that requires subjects to cross out as quickly as possible target pictures within a large array of target and nontarget pictures. For each item of the Nonverbal Stroop subtest, subjects are presented with a target stimulus picture (consisting of two colored circles, one of which has a white dot in the center) and must identify as quickly as possible a response picture (from four possible choices) that exactly matches the target stimulus picture. Only the Color Incongruent section (the two circles of the target stimulus are different colors) of the Nonverbal Stroop was used.

Parent-reported executive functioning of the child's everyday behaviors was assessed using subscales from two questionnaire measures: the Learning, Executive, and Attention Functioning Scale (LEAF) (Castellanos, Kronenberger, & Pisoni, 2018) and the Behavior Rating Inventory of Executive Function, Second Edition (BRIEF-2) (Gioia, Isquith, Guy, & Kenworthy, 2015). Three subscales from the LEAF (Attention, Working Memory, and Processing Speed) and three subscales from the BRIEF (Inhibit, Shift, and Working Memory) were selected to correspond to core executive functioning domains, particularly those identified as being at-risk in children and adolescents with CIs (Kronenberger & Pisoni, 2018). LEAF subscale raw scores (the LEAF is criterion-referenced and does not have norms) and BRIEF subscale T-scores (age- and gender-based norms) were used to assess executive functioning in everyday behavior.

Data Analysis Approach

In order to evaluate the reliability and subscale structure of the QHS, internal consistency (alpha) and corrected item-to-total correlations were calculated for the four subscales and 21 items. Subscale scores were then factor analyzed (principal axis factoring) to investigate the higher-order factor structure of the QHS (e.g., Total score(s)). Next, in order to evaluate differences between the hearing groups on parent-reported functional everyday hearing, all 21 QHS items were compared between the CI and NH samples using a MANOVA, with follow-up t-tests for individual items, subscales, and total scores using t-tests if the

MANOVA was statistically significant. This analysis also provided a methodology to assess the construct validity of the QHS, since children with hearing loss would be expected to score lower on a measure of functional everyday hearing than normal-hearing peers. Finally, Pearson correlations were calculated separately for the CI and NH samples between QHS scores and measures of audiological functioning, hearing history, speech perception, language, and executive functioning. In order to reduce potential alpha error, these correlations were calculated first using only the QHS Total score. If the correlation between the QHS Total score and any specific measure of audiological functioning, hearing history, speech perception, language, or executive functioning was statistically significant ($p<0.05$), additional correlations for the QHS subscales and that specific measure were then calculated.

Results

Sample Characteristics

The samples did not differ in age, gender, or household income (Table 1). Both samples scored above the normative scaled score mean of 10 on the measure of nonverbal intelligence (Leiter-3 Classification and Analogies subscale), and the NH group scored significantly higher on that measure than the CI group. The CI sample had an average age at implantation of 21.1 months and had used their CIs, on average, for 10.6 years (Table 1). The CI sample showed poorer functioning on all measures of speech perception, language, working memory, and parent-reported executive functioning than the NH sample, while the groups did not differ significantly on the Leiter-3 measures of Attention Sustained and Nonverbal Stroop (Table 3)

Quality of Hearing Scale: Subscale Derivation and Comparison of CI and NH Samples

QHS subscales demonstrated excellent internal consistency (IC) reliability in the combined sample of CI users and NH peers, with alpha values ranging from 0.90 to 0.96 (Table 2). Corrected item-to-total correlations for items within subscales exceeded 0.6 for all items and exceeded 0.7 for 19/21 items, indicating that all items strongly reflected their constituent subscale constructs. In order to derive composite/total score(s), scores on the four subscales were factor analyzed using principal axis factoring. Eigenvalues for the factors were 3.27, 0.27, 0.24, and 0.21, which strongly supported a single factor underlying the four subscale scores.

As hypothesized, the NH sample scored higher than the CI sample (indicating better functional hearing) the QHS Total score and on all subscale and item scores of the Speech, Localization, and Sounds subscales (Table 2; MANOVA comparing CI and NH for all items, $F(21,64)=6.44$, $p<0.001$). In the CI sample, functional hearing in noisy background situations (e.g., QHS items 2-6, 7, 10-12, 13-14, and 16) was typically rated as poorer (raw scores in the 5.0-7.6 range) than functional hearing in quiet situations (e.g., QHS items 1, 8, and 15; raw scores in the 7.0-9.4 range). The mean scores for the Speech and Sounds subscales of the QHS in the CI sample fell in a descriptive range between "Pretty Much" (the anchor for a score of 6) and "Very Much" (the anchor for a score of 8), indicating good functional hearing (albeit lower than that for the NH sample). The CI sample scored higher

on the Effort subscale than the NH sample, indicating the use of more listening effort during hearing.

Association of Quality of Hearing Scale Scores with Audiological/Hearing History, Speech Perception, Language, and Executive Functioning Scores

QHS Total scores correlated significantly and positively with income level in the CI sample and with chronological age in the NH sample (Table 4). Further analyses with QHS subscales for these variables showed that better QHS Speech, Localization, and Sounds subscale scores correlated with higher income level in the CI sample (Table 5) and that higher QHS Effort subscale scores (e.g., more effort) were associated with younger ages in the NH sample (Table 6). The QHS Total score was not significantly associated with any of the audiological or hearing history variables in the CI sample. However, correlations of QHS Total scores with age at implantation and duration of deafness approached significance (p) < .10), with earlier implantation and shorter duration of deafness associated with better QHS Total scores. Furthermore, although bilateral vs. unilateral CI users did not differ significantly on the QHS Total score (t(40)=0.24, $p = 0.81$), bilateral CI users had significantly better QHS Localization subscale scores than unilateral users (t(40)=2.36, p < 0.03).

QHS Total scores were significantly positively correlated with three of the four speech perception measures in the CI sample (CNREP and HINT-C sentences in quiet and noise) (Table 4). Follow-up correlational analyses with QHS subscales in the CI sample (Table 5) indicated that participants with better Speech and Sounds subscale scores had better speech perception scores, while localization and effort scores were less consistently related to speech perception.

For the examiner-administered tests of executive functioning in the CI sample, better QHS Total scores were associated with better performance on verbal working memory tests (VDS and LNS) but were uncorrelated with examiner-administered nonverbal tests of inhibitionconcentration and controlled fluency-speed (Attention Sustained and Nonverbal Stroop). Higher QHS Speech and Sounds subscale scores and lower QHS Effort scores (e.g., lower listening effort needed in functional hearing settings) were associated with better performance on the verbal working memory tests in the CI sample (Table 5). For the NH sample, only LNS was significantly correlated (positively) with QHS Total scores (Table 4), as a result of significant correlations of LNS scores with the QHS Speech (positive) and QHS Effort (negative) scores (Table 6). No other examiner-administered tests of executive functioning were correlated with QHS scores in the NH group.

Greater problems in multiple domains of executive functioning (by parent report on the LEAF and BRIEF-2 Attention, Processing Speed, Shift, and Working Memory subscales) were consistently related to poorer functional hearing quality (higher QHS Total scores) in the CI sample (Table 4). Correlations between QHS subscales and LEAF/BRIEF-2 subscales indicated that higher scores on the Speech and Sounds subscales and lower scores on the Effort subscale were related to fewer problems in executive functioning in the CI sample. The pattern of correlations for QHS Total score with LEAF/BRIEF-2 subscales in the NH

sample mirrored the results of the CI sample, but only for Attention and Working Memory (not for Shift or Processing Speed).

Discussion

This study investigated associations between functional everyday hearing and audiological, speech perception, language, and executive functioning in samples of children and adolescents with CIs or NH, using a new parent-report measure of functional hearing quality (the Quality of Hearing Scale – QHS). Results supported the internal consistency of four QHS subscales (Speech, Localization, Sounds, and Effort) as well as a composite score (QHS Total score). All QHS items, subscales, and the total score indicated poorer functional everyday hearing quality in CI users compared to NH peers, although mean QHS scores suggested that much of the time CI users were able to adequately hear (detect and discriminate), recognize, and understand speech and sounds. QHS Total scores were not significantly correlated with audiological measures, hearing history, or nonverbal intelligence in CI users, but were significantly correlated with almost all measures of speech perception, sentence recognition, language, verbal working memory, and executive functioning in CI users. In contrast, QHS scores were not correlated with speech perception or language scores in the NH sample and correlated less consistently with verbal working memory and executive functioning in the NH sample compared to the CI sample.

Psychometric (internal consistency and factor) analyses supported the composition of the QHS into four subscales and a Total score, all of which differentiated children with CIs from those with NH. Post-hoc analyses showed that the four QHS subscales were related but distinct in both samples: In the NH sample, intercorrelations among QHS subscales ranged from 0.53 for the Speech and Sounds Subscales to 0.81 for the Localize and Sounds subscales, with all other intercorrelations in a 0.57-0.59 range. In the CI sample, intercorrelations ranged from 0.59 for the Localize and Effort subscales to 0.71 for the Speech and Sounds subscales, with all other intercorrelations in a 0.64 to 0.69 range. Thus, with the exception of the Localize and Sounds subscales in the NH sample, all QHS subscales showed between 28% and 50% shared variance, demonstrating both overlap and statistical independence of the subscales.

The finding of better functional everyday hearing in NH children compared to children with CIs supports the validity as well as the clinical utility of the QHS items and subscales. Stronger functional hearing quality in NH children relative to children with CIs is expected given that CIs do not fully encode and reproduce the richness and fine acoustic-phonetic details of speech sounds when compared to typical acoustical hearing (representational specificity) (Wilson & Dorman, 2008). Thus, differences between CI and NH samples on all QHS item, subscale, and total scores support the construct validity of the QHS as a measure of functional hearing quality.

Within the CI sample, QHS Total scores were not significantly associated with measures of audiological functioning and hearing history. However, the ranges on some of these variables were restricted by study inclusion criteria such as deafness prior to age 3 years and implantation at age 3 years, 11 months and younger. Greater variability in scores on these

variables may reveal stronger associations with QHS scores. For example, a statistical trend $(p < .10)$ was found for earlier age at implantation and shorter duration of deafness to be correlated with better functional hearing (QHS Total). Furthermore, bilateral CI users displayed significantly better QHS Localization subscale scores than unilateral CI users. Thus, QHS scores may show relations with some selective components of audiological functioning and hearing history, although the magnitude of these relations was small in this sample. This finding demonstrates that functional hearing in CI users is not substantially captured by traditional audiometric or hearing history measures and must be assessed as a separate audiological outcome measure following implantation.

QHS scores were significantly correlated with nonword repetition and sentence recognition scores (in quiet and noise) in the CI sample, particularly for the Speech and Sounds subscales. This finding provides validation for the QHS as a measure of functional hearing in everyday life, by showing that ratings of better hearing of speech/sounds on the QHS are found for children with stronger speech perception and sentence recognition skills as measured by widely-accepted, standardized, individually-administered tests. The QHS Effort score correlated significantly only with the nonword repetition test score, with greater functional listening effort associated with poorer nonword repetition test scores. This finding is consistent with previous theory and empirical research suggesting that children with CIs who have poorer fast-automatic rapid phonological coding skills (assessed by nonword repetition test performance) must expend greater compensatory effort during spoken language processing (Pichora-Fuller et al., 2016; Smith, Pisoni, & Kronenberger, 2019). LNT scores did not correlate significantly with the QHS Total score, and post-hoc correlations using separate scores for lexically-easy and lexically-hard words with QHS Total scores were also not statistically significant. This finding may reflect the less cognitively challenging nature of speech perception on the LNT, as compared to the nonword repetition and HINT sentences, both of which place greater demands on phonological/lexical memory as a result of greater length/complexity than the simple, familiar words of the LNT.

Both language test scores (PPVT-4 and CELF-5 Following Directions) correlated significantly with QHS Total, Speech, Sounds, and Effort scores in the CI sample. Although the correlational nature of this study makes it impossible to establish the direction of effects between functional hearing and language, these effects may be reciprocal, with better hearing contributing to better language (as a result of greater auditory experience) and better language skills contributing to more efficient processing of spoken auditory information. Importantly, hearing effort on the QHS was also related to language ability, with greater effort associated with poorer language ability. This finding is consistent with theories and empirical evidence suggesting that stronger language skills support more efficient spoken language processing, requiring less effort during language processing (Rönnberg et al., 2013).

In contrast to the strong associations found between functional hearing, speech perception, and language skills in CI users, functional hearing quality on the QHS was unrelated to speech perception and language skills in NH peers. It is likely that the strong functional hearing skills of the NH sample (e.g., mean QHS Total and Subscale scores were 8.9-9.2 on

a 1-10 scale) supported speech perception and language at a more uniform, high level in the NH sample, rendering functional hearing less important for explaining variability in speech and language outcomes in NH youth. A high level of functional hearing quality (albeit with some variability, as indicated by standard deviations of approximately 1 on QHS subscales in the NH sample) would be expected in the NH sample based on study entry criteria, which required passing a hearing screening and parent-report of no significant past or current hearing problems. Thus, this finding further supports the construct validity of the QHS.

On measures of executive functioning that were individually-administered in the office setting (VDS, LNS, and Leiter-3 subscales), QHS scores correlated significantly only with measures of verbal working memory in the CI sample (VDS and LNS), but not with nonverbal measures of controlled fluency-speed/inhibition-concentration (Leiter-3 subscales). Because the digit span test was administered visually, the correlation between functional hearing quality and VDS performance does not reflect a direct effect of hearing and audibility during the test administration. Rather, this correlation may reflect the impact of hearing quality on the quality of underspecified/coarse-coded phonological/lexical representations of words in short-term memory during this working memory task. Because the LNS test was administered in spoken format (with spoken response), its correlation with functional hearing quality may be the result of hearing problems during that test administration or the result of underspecified phonological/lexical representations of words in memory.

QHS-assessed functional hearing quality in the NH sample correlated significantly only with the VDS Backward score. VDS Backward places additional concurrent demands on processing during short-term verbal memory (e.g., reversing digits concurrently with holding them in short-term memory) and is typically considered to be a more classic "working memory" test (Alloway, 2007). Thus, the correlation between functional hearing quality and VDS Backward in the NH sample may reflect a stronger association between functional hearing and a more challenging test of working memory compared to VDS Forward. For CI users, on the other hand, forward digit span tasks are much more demanding than those tasks are for NH peers, as a result of additional demands on speech perception and storage and maintenance of coarsely-coded underspecified phonological and lexical representations of spoken words in memory (Kronenberger & Pisoni, 2018); thus, VDS forward in the CI sample may reflect a stronger working memory component than in the NH sample, explaining the association of functional hearing and VDS forward in the CI sample but not in the NH sample.

The Attention Sustained and Nonverbal Stroop subtests of the Leiter-3 were unrelated to functional hearing in either (NH or CI) sample. This may reflect a modality-specific effect, with functional hearing impacting verbally-based executive functions more than visuallybased executive functions. Alternatively, functional hearing quality might not be as important for inhibition-concentration and controlled fluency-speed dimensions of executive functioning. A third possibility is that the Attention Sustained and Nonverbal Stroop tests might be less valid measures of executive functioning than the working memory tests. Further investigation of the association of functional hearing quality with additional measures of executive functioning is recommended to better understand this finding.

In the CI sample, QHS Total, Speech, Sounds, and Effort scores were consistently and strongly related to all parent-report questionnaire measures of EF, with the exception of the BRIEF-2 Inhibit subscale. Furthermore, correlations between the QHS Effort score and parent-reported executive functioning were large (in the range of 0.41-0.53), consistent with models and empirical research demonstrating that executive functioning is recruited to initiate, direct, and control mental effort, including effort allocated to hearing and spoken language (Kronenberger, Henning, Ditmars, & Pisoni, 2018). Furthermore, strong associations between functional hearing quality and executive functioning are consistent with theories positing that auditory experience influences the development of executive functioning, particularly in children with early hearing loss (Kronenberger & Pisoni, 2020). Some prior research has not found associations between audiological measures, hearing history, and executive functioning (Kronenberger, Colson, Henning, & Pisoni, 2014; Kronenberger et al., 2013), but that research did not investigate relations of functional hearing quality and executive functioning.

Post-hoc analyses demonstrated that the significant correlations between functional hearing quality and LEAF/BRIEF-2-assessed executive functioning reflect direct associations between hearing and executive functioning that are not fully explained or mediated by the association of hearing and language: Even with PPVT-4 and CELF-5 Following Directions scores statistically controlled, partial correlations between QHS Total scores and all LEAF/ BRIEF-2 subscales in this study remained significant with the exception of LEAF Attention (r=−0.27, p < .10). Furthermore, partial correlations for the QHS Effort score were significant for all LEAF/BRIEF-2 subscales in the CI sample with the language tests statistically controlled (a table of all partial correlations is available from the authors). Thus, the association between functional hearing quality and executive functioning was not solely mediated by language. The causal direction of this association, however, is unclear from these correlational results. Bidirectional effects may underlie the significant correlation between functional hearing quality and executive functioning, with hearing experience contributing to the development of executive functioning and executive functioning supporting hearing quality through the direction and allocation of effort (Kronenberger & Pisoni, 2020).

Correlations between functional hearing quality and parent-reported measures of executive functioning in the NH sample were less robust and consistent than in the CI sample. Working Memory and Attention subscale scores were significantly related to functional hearing quality in the NH sample. However, LEAF and BRIEF-2 subscales assessing controlled fluency-speed, flexibility-shifting, and inhibition were not significantly correlated with functional hearing quality in the NH sample. These findings suggest that cognitive control processes specifically under attention and working memory demands (but not those controlling speed, flexibility, or inhibition) are strongly associated with functional hearing quality in youth with NH.

Analyses of the QHS subscales in the CI sample indicated that quality of hearing scores for speech and sounds were most strongly related to speech, language, and executive functioning outcomes. Hearing effort, on the other hand, was less strongly related to speech perception and more strongly related to language and executive functioning. Prior research

has demonstrated that speech perception may be more dependent on fast-automatic processing abilities (reflected by functional hearing quality for speech and sounds) than on slow-effortful processing (Smith et al., 2019), although the importance of slow-effortful processing is greater when fast-automatic processing is not effective (Rönnberg et al., 2013). Language and executive functioning, on the other hand, demand more higher-order, purposeful, directed, cognitive processing, which requires greater recruitment of sloweffortful processing strategies (Kronenberger et al., 2018).

Contrary to findings with the other QHS subscales, the QHS Localization subscale was minimally related to almost all measures of speech perception, language, and executive functioning. Compared to functional hearing quality for speech, sounds, and effort (which allow for better speech perception, more language experience, and greater deployment of executive functioning), localizing sounds in space is less directly relevant for the development of speech perception, language, and executive functioning skills. However, localization of sound is an important component of auditory functioning in the everyday environment because it facilitates orientation to and identification of auditory stimuli (Gordon, Jiwani, & Papsin, 2013; Litovsky & Gordon, 2016). Importantly, CI users with bilateral implants showed better QHS Localization scores than CI users with unilateral implants, consistent with prior research demonstrating the advantage of bilateral over unilateral CIs for localization of sound in space (Gordon et al., 2013; Litovsky & Gordon, 2016). Furthermore, this difference between bilateral and unilateral users on the Localization subscale remained statistically significant with age statistically controlled in an analysis of covariance (F(1,39)=5.7, $p < .03$). Nevertheless, because the Localization subscale was not validated against a formal test of localization performance, additional investigation of the validity of the Localization subscale is recommended.

The findings of this study must be considered in the context of several limitations. First, study results are correlational and therefore cannot definitively establish direction of causality. Second, the audiological measures in the CI sample had limited ranges due to study entry criteria and were not available for some subjects as a result of incomplete or inaccessible medical records. Third, the NH group provided only limited speech perception data as a result of ceiling effects that would have occurred for the conventional administration procedures for the LNT and HINT-C; future research may use degraded stimuli or background noise to provide a more challenging test of speech perception for NH listeners.

Although multiple statistical tests were conducted to evaluate the research questions of this study (raising the risk for alpha error), several processes were also used to reduce alpha error: (1) a global statistically significant MANOVA test for QHS items was required prior to analyzing specific items and subscales; (2) a statistically significant correlation between the QHS Total score and each measure of audiological, hearing history, speech perception, language, and executive functioning was required prior to investigating correlations between QHS subscales and that measure; and (3) patterns of significant correlations were emphasized over isolated statistically significant correlational results.

Finally, because the QHS is a questionnaire based on parent-report, rater issues including bias, lack of awareness, misunderstanding item content, and social desirability may have affected QHS scores. For example, if parents are not aware of some components of their child's listening experience, they may answer QHS items inaccurately. Furthermore, because parents completed the executive functioning questionnaires in addition to the QHS, method bias (shared variance in questionnaires as a result of having the same rater) may have influenced results. On the other hand, significant correlations between QHS scores and speech, language, and neurocognitive functioning scores argue strongly for the validity of the QHS as a clinically useful measure of functional hearing quality. Although our focus in this paper was on parent-reported quality of hearing across the broad childhood age range, a self-report questionnaire version of the QHS for adolescents and young adults would address some of these limitations and provide additional converging information about functional hearing quality.

The findings of this study have several clinical implications for the assessment of hearing outcomes in children and adolescents with CIs, as well as for understanding and explaining the enormous variability and individual differences in speech, language, and neurocognitive outcomes in this clinical population. First, results demonstrated that functional hearing quality can be validly assessed in a parent-report questionnaire format and that functional hearing quality is important for understanding hearing outcomes above and beyond results obtained from conventional audiometric measures. As a result, assessment of functional hearing outcomes should routinely be performed as a part of audiological evaluations for children and adolescents with CIs. Second, because functional hearing quality is associated with speech, language, and executive functioning outcomes, functional hearing quality should be assessed as an outcome that is relevant for determination of CI benefit and effectiveness. Third, because a questionnaire can be used to assess functional hearing quality based on responses provided by parents, evaluation of functional hearing quality can be completed quickly, inexpensively, and with little direct burden on the child. As a result of these data-driven clinical implications, it is recommended that questionnaire-based assessment of functional hearing quality be considered as a core component of audiological outcome measurement after cochlear implantation.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Table 1.

Sample Demographics and Hearing History

Note: CI=cochlear implant; SD=standard deviation; NA=not applicable; degrees of freedom (df) for t-tests=84; p value for gender obtained from a Fisher's exact test, 2-sided.

 \int_a^a in Years;

 b _{in} Months;

 $c_{PTA=pure}$ tone average for frequencies 500, 1000, 2000 Hz in dB HL;

 $d_{\text{Communication mode coded mostly sign (1) to auditory-verbal (6) (Geers & Brenner, 2003);}$

 e^e On a 1 (under \$20,000) to 8 (\$150,000+) scale;

f Leiter-3 Classification and Analogies (scaled score).

 $p < .05$

Table 2.

Quality of Hearing Scale (QHS) Items and Subscales

Note: CI=cochlear implant; NH=normal-hearing; IC=internal consistency (Cronbach's alpha for subscales and corrected item-to-total correlations for items). Values for CI and NH samples are Mean (SD). All t-tests (df=84) were statistically significant at $p < .001$, except Items 2 and 17 (p < .01). Subscale scores are means of constituent item scores. "QHS Total Score" was obtained by taking the mean of the four subscales, with the Effort subscale reverse-scored.

Table 3.

Sample Speech Perception, Language, and Executive Functioning

Note: CI=cochlear implant; NH=normal-hearing. CELF=Clinical Evaluation of Language Fundamentals; LEAF=Learning, Executive, and Attention Functioning Scale; BRIEF-2=Behavior Rating Inventory of Executive Function. df=83 for Speech Perception, Language, and Individually-Administered Executive Functioning measures with the exception of Nonverbal Stroop (df=82); df=84 for Parent-Report measures.

** $p < .01;$

 $p < .001$

Table 4.

Quality of Hearing Scale Total Score Correlations with Outcome Measures

Note: Values are Pearson correlations; CI=cochlear implant;

 a PTA=pure-tone average for frequencies 500, 1000, 2000 Hz in dB HL;

b
Communication mode coded mostly sign (1) to auditory-verbal (6) (Geers & Brenner, 2003);

 $c_{\text{On a 1 (under $20,000)$ to 8 ($150,000+)$ scale}}$

d Leiter-3 Classification and Analogies (scaled score).

CELF=Clinical Evaluation of Language Fundamentals; LEAF=Learning, Executive, and Attention Functioning Scale; BRIEF-2=Behavior Rating Inventory of Executive Function.

$$
a_{p<.10)}
$$

 p^* = .05;

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Table 5.

Quality of Hearing Scale Subscale Correlations with Outcomes in Cochlear Implant Sample

Note: Values are Pearson correlations; Local=Localization subscale; Income Level coded on a 1 (under \$20,000) to 8 (\$150,000+) scale; CELF=Clinical Evaluation of Language Fundamentals; LEAF=Learning, Executive, and Attention Functioning Scale; BRIEF-2=Behavior Rating Inventory of Executive Function.

 $^{a}p<.10;$

 $p < .05;$

**
 $p < .01$

 $p < .001$

Table 6.

Quality of Hearing Scale Subscale Correlations with Outcomes in Normal Hearing Sample

Note: Values are Pearson correlations; Local=Localization subscale; LEAF=Learning, Executive, and Attention Functioning Scale; BRIEF-2=Behavior Rating Inventory of Executive Function.

 $^{a}p<.10;$

* $p < .05;$

** $p < .01$