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Comparisons of IQ in Children with and without Cochlear Implants: Longitudinal Findings and Associations with Language

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Abstract

Objectives—To make longitudinal comparisons of intelligence (IQ) in children with cochlear implants (CIs) and typical hearing peers from early in development to the school-age period. Children with additional comorbidities and CIs were also evaluated. To estimate the impact of socioeconomic status and oral language on school-age cognitive performance.

Design—This longitudinal study evaluated nonverbal IQ in a multicenter, national sample of 147 children with CIs and 75 typically hearing peers. IQ was evaluated at baseline, prior to cochlear implantation, using the Bayley Scales of Infant and Toddler Development and the Leiter International Performance Scale. School-age IQ was assessed using the Wechsler Intelligence Scales for Children. For the current study, only the Perceptual Reasoning and Processing Speed indices were administered. Oral language was evaluated using the Comprehensive Assessment of Spoken Language.

Results—Children in the CI group scored within the normal range of intelligence at both time points. However, children with additional comorbidities scored significantly worse on the Processing Speed, but not the Perceptual Reasoning Index. Maternal education and language were significantly related to school-age IQ in both groups. Importantly, language was the strongest predictor of intellectual functioning in both children with CIs and normal hearing.

Conclusion—These results suggest that children using cochlear implants perform similarly to hearing peers on measures of intelligence, but those with severe comorbidities are at-risk for cognitive deficits. Despite the strong link between socioeconomic status and intelligence, this association was no longer significant once spoken language performance was accounted for. These

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results reveal the important contributions that early intervention programs, which emphasize language and parent training, contribute to cognitive functioning in school-age children with CIs. For families from economically disadvantaged backgrounds, who are at-risk for suboptimal outcomes, these early intervention programs are critical to improve overall functioning.

Introduction

To date, few studies have measured intelligence in severely to profoundly deaf children with cochlear implants (CIs). Traditionally, outcomes for children with CIs have focused on speech perception, oral language skills, and literacy while ignoring the important role cognition plays in facilitating these outcomes (Blamey et al. 2000; Pyman et al. 2000; Sarant et al. 2001; Geers et al. 2003; Marschark et al. 2007; Quittner et al. 2007; Niparko et al. 2010; Knoors & Marschark 2014). Despite the known benefits of cochlear implantation, there is well-documented variability in these outcomes (Svirsky et al. 2000; Niparko et al. 2010; Geers & Sedey 2011) that may be related to cognitive abilities (e.g., visual attention) (Smith et al. 1998; Cejas et al. 2015; Quittner et al. 2016). Measures of intelligence are the most broad-based, valid indicators of these developing abilities in children, and are significantly related to language learning, behavioral and social development, and academic functioning (Marschark et al. 2007; Barker et al. 2009; Wiefferink et al. 2012; Hoffman et al. 2014, 2015; Quittner et al. 2016). In children with CIs, cognition has been shown to be a strong predictor of oral language (Sarant et al. 2001; Geers et al. 2003).

Although a few studies have examined intelligence in children with CIs, they have been limited by small samples, lack of racial and ethnic diversity, and later implantation (age greater than 3 years) (Schlumberger et al. 2004; Khan et al. 2005). The purpose of this study was to examine intelligence quotients (IQ) in a large, national multi-center study of children implanted, at approximately age two, and their hearing peers. This longitudinal study enabled us to assess IQ in children pre-implant and again during the school-age period. Further, we examined relationships between IQ and oral language development several years after implantation.

Previous IQ studies in children with CIs

Importantly, studies of intelligence in children with hearing loss should utilize nonverbal measures. Nonverbal intelligence tests were designed to measure general cognition without the confound of language ability. In addition, these measures eliminate the need for verbal instructions throughout administration. There are nonverbal IQ measures, such as the Leiter International Performance Scale (Roid & Miller 1997), which were designed specifically for children with hearing loss or alternatively, the Perceptual Reasoning and Processing Speed Indices from the Wechsler Intelligence Scale (WISC-IV) (Wechsler 2003). These measures typically include tests of visual-spatial skills, processing speed, and fluid intelligence (e.g., finding patterns and relationships in novel problems). Results of prior studies measuring IQ in this population will be reviewed based on the age of the child, from the youngest to oldest.

Khan, Edwards, and Langdon (2005) assessed nonverbal intelligence using the Leiter-R in 25 preschoolers with CIs who were implanted at about age 3 (mean age at IQ assessment =

4.22), 13 preschoolers with hearing aids (moderately-severe-to-profound hearing loss), and 18 hearing peers. This IQ assessment was performed one year following implantation for those in the CI group. Children with CIs scored significantly better than the hearing aid group on Full Scale intelligence, the Fluid Reasoning composite, and three subtests: Sequential Order (measures rule generation based on sequential information), Associated Pairs (measures associative memory), and Forward Memory (similar to a digit span task). However, it is important to note that both the CI and hearing groups came from families with higher socioeconomic status and both of these groups scored above average on Full Scale IQ. When compared to the hearing group, children with CIs scored similarly on Full Scale intelligence and all subtests; however, they evidenced worse performance on the Attention Sustained subtest. These findings of poor visual sustained attention are consistent with other studies reporting delays in attention in young children with CIs (Quittner et al. 1994; Smith et al. 1998; Horn et al. 2005; Quittner et al. 2007; Barker et al. 2009).

In a study of young, school-age children, ages 5 to 9 years with hearing aids ($n=29$), CIs ($n=25$), and normal hearing ($n=40$), Schlumberger and colleagues (2004) compared nonverbal intelligence across these groups. Median age at implantation was 26 months, with a range of 12 to 39 months. They administered the Raven's Standard Progressive Matrices, which measures non-verbal and fluid intelligence, as well as visual spatial tasks. As in the previous study, children with CIs performed similarly to the hearing controls and better than the hearing aid group on the Raven's Matrices, a copy drawing task, and a perceptual reasoning task (Mazes subtest from the WISC-III). The authors assumed that children with hearing aids, who had severe-to-profound losses, had worse auditory input than the children with CIs and thus, scored lower than both other groups. However, it should be noted that the hearing aid group had more children from low SES backgrounds than the CI or hearing group, and this confounding variable may partially explain the results. The authors argued that children received high quality special education resources and, therefore, any effects of SES on IQ were minimal. They also compared outcomes on the Raven's Matrices based on educational status of the parents and found no differences across groups; thus, they attributed differences in IQ to poor auditory input. However, the influence of SES on cognitive development is well-established and may be an alternative explanation for these results (Hanscombe et al. 2012). One aim of our study was to examine the influence of SES on IQ during the school-age period.

Similar to Khan et al. (2005), De Giacomo and colleagues (2013) assessed IQ using the Leiter-R in an older sample of children with unilateral implants ($n=20$; mean age at assessment = 9.17 years) versus hearing peers ($n=20$). The average age at implantation for these children was 3.13 years. First, despite both groups scoring, on average, below the mean on IQ, there were no differences in IQ between the CI and hearing groups. The mean IQ for the CI group was 89.05 ($SD=13.68$) compared to 90.10 ($SD=21.85$) in the hearing controls. Second, results showed that 55% of children with CIs scored in the average range (intelligence ≥ 85) on Full Scale IQ, 40% in the borderline range, and 5% in the mild impairment range. However, they did not examine performance on any composite scales or individual subtests and provided no information on SES or hearing history. Finally, age at implantation was negatively associated with IQ scores, which indicated that earlier

implantation facilitated cognitive development. This study was limited by a small sample size and the low performance of the hearing peers.

In the largest study to date evaluating intelligence in 112 school-age children with CIs, IQ was measured at ages 8–9 and again at 15–18 years (mean age at implantation = 3.6 years) (Geers et al. 2011). Nonverbal intelligence was measured using 3 subtests from the WISC-III: Picture Completion, Picture Arrangement, and Block Design. Assessments of IQ at each time point fell within the average range (Performance IQ at ages 8–9 = 103.2, $SD = 14.0$; Performance IQ during high school = 103.1, $SD = 16.0$). IQ scores predicted both academic outcomes and language and reading abilities in high school. Family SES, which was measured using ratings of family income and parental education level, was significantly related to all outcome variables. These findings suggested that nonverbal IQ is related to academic performance and the WISC is appropriate for use with deaf children. Strengths of this study include the relatively large sample size and longitudinal design, which validated the stability of IQ scores measured in school-age children. In contrast, we examined longitudinal associations between IQ measured pre-implantation (mean age 2 years) and post-implantation (mean age 8 years).

Relationships between IQ and Oral Language

Nonverbal IQ is one of the most robust predictors of positive language outcomes in children with CIs (Phillips, Wiley, Barnar, & Meinzen-Derr, 2014). To date, several theories aim to explain the interrelationship between cognition and language. The Cattell-Horn-Carroll theory postulates that nonverbal IQ measures as a whole draw heavily on visual processing abilities and fluid intelligence (DeThorne & Schaefer, 2004). Fluid intelligence refers to “mental operations that an individual may use when faced with a relatively novel task that cannot be performed automatically.” Moreover, visual processing speed is the ability to generate, analyze, manipulate, and think with visual patterns and stimuli. Research has reported that not only are verbal IQ and performance IQ moderately correlated in children and adults, but there is also evidence that verbal processing speed predicts later language development. This provides evidence that nonverbal IQ taps into the global factor of intelligence. Other studies have reported that once children are old enough to form and process utterances (18–30 months), limitations on working memory will affect comprehension, production and learning (Deak, 2014). Thus, there are clear linkages between cognition, working memory, and language abilities.

Geers, Nicolas, and Sedey (2003) examined the language skills of 181 children ages 8–9 (mean age at implantation = 3.42 years) and found that nonverbal IQ, measured using the Performance Index of the WISC-III, predicted expressive language skills. Furthermore, nonverbal IQ predicted total language, which combined both speech and signing capabilities. Similarly, in a different study using the same sample, Tobey and colleagues (2003) found that nonverbal IQ was significantly related to speech intelligibility scores. Other studies have also linked specific domains of cognitive abilities, such as working memory, to strong expressive language skills (Cleary et al. 2001).

In addition to expressive language, nonverbal IQ has been shown to predict receptive language abilities. Geers, Nicolas, and Moog (2007) evaluated predictors of receptive

language in a sample of 126 children with CIs, who were assessed between the ages of 5–6 years (age at implantation = 11 to 59 months). Nonverbal IQ individually accounted for 10% of the variance in receptive language, and was related to vocabulary scores and parental education. It was noted that children with high IQs received their CIs at younger ages, which may have also boosted cognitive development. Specific domains of nonverbal IQ have been related to receptive language outcomes, as well. For example, Dawson and colleagues (2002) found that visual spatial memory predicted receptive language skills in 24 school-age children with CIs. Finally, nonverbal IQ has also been related to strong reading skills (Geers et al. 2003).

Thus, nonverbal IQ scores are related to expressive and receptive language skills and academic outcomes in children with CIs and may serve as one of the best predictors of positive outcomes following cochlear implantation. However, it is unclear how IQ scores pre-implant relate to later cognitive abilities. This study will be the first to examine IQ prior to and following implantation.

In sum, previous research has yielded consistent findings of similar nonverbal IQ when comparing children with CIs to their hearing peers at various ages. Furthermore, IQ has been shown to be positively related to oral language, reading abilities, and academic performance. However, several issues must be considered. First, several previous studies have utilized small, single-site samples, which lacked racial and ethnic diversity. In addition, little attention has been given to evaluation of cognitive functioning in children with CIs who have additional comorbidities (Cruz et al. 2012). Second, several studies failed to control for age at implantation, which has been cited as a concern (Marschark et al. 2007). Third, findings regarding the effects of family factors on IQ, such as SES were mixed, despite the recognition of their impact on education and academic performance (Uziel et al. 2007; Geers et al. 2008). Finally, the majority of previous studies had children with an average age at implantation of 3 years or older, which does not reflect current clinical practice.

In contrast, the present study was based on a large sample of children with CIs recruited from multiple centers across the United States, specifically chosen to maximize diversity (Wang, et al. 2012). Furthermore, this study evaluated factors related to cognitive abilities, including age at implantation, gender, parental education, and income. Children included were also implanted at a younger age (mean age at implantation = 2.4 years), which is more representative of those currently receiving CIs. Finally, this is the only study to date that measured intelligence both prior to implantation and again during the school-age period, facilitating longitudinal analyses.

The major aim of this study was to make longitudinal comparisons between children with CIs and hearing peers. Comparisons of IQ scores were made across three groups: children with CIs, children with CIs + Comorbidities, and typically hearing children. Given the prior literature, we expected that the CI and typically hearing children would perform within the normal range on IQ assessments. Next, we hypothesized that children with comorbidities would perform significantly worse than those without comorbidities. Note that these comparisons were exploratory given the small sample size. A secondary aim was to examine the associations between IQ scores from early childhood, pre-implantation, to school-age

performance post-implantation. We expected the baseline and school-age IQ scores to be correlated. Third, we evaluated the associations between IQ scores in school-age children and language ability. We hypothesized that IQ and language scores would be correlated. Finally, we tested a multiple regression model across the CI and hearing groups to evaluate predictors of school-age IQ.

Materials and Methods

Participants

Data were drawn from the Childhood Development after Cochlear Implantation (CDaCI; NIDCD R01DC004797) study, which is a multi-center, national cohort investigation of the effectiveness of CIs in deaf children in relation to their hearing peers (Fink et al. 2007). The larger parent study examined a variety of outcomes in children before and after cochlear implantation, including expressive and receptive language, quality of parent-child interactions, joint attention, psychosocial development, and health-related quality of life (Niparko et al. 2010; Cruz et al. 2013; Quittner et al. 2013; Cejas et al. 2015).

Participants were recruited from 6 CI centers and 2 preschools that enrolled children with and without hearing loss. Inclusion criteria for deaf children were: 1) age under 5 years, 2) severe-to-profound sensorineural hearing loss, and 3) a commitment to raise the child using spoken English. Participants who scored within the severely delayed range on a cognitive screener using the Bayley Psychomotor Index (Bayley 1993) or the Leiter International Performance Scale – Revised (Roid & Miller 1997) were excluded. Children of a similar age with normal hearing were also enrolled. A total of 188 children with CIs (mean age at baseline = 2.2 years) and 97 hearing peers (mean age at baseline = 2.3 years) were enrolled. See Fink et al. (2007) for a more detailed description of the sample. All participants were assessed at baseline (prior to implantation for those in the CI group) and every 6 months for 3 years. Following that, participants were assessed annually for the next 5 years. This study was approved by the Institutional Review Boards at all sites and all parents gave written, informed consent and children gave assent, when appropriate, prior to completing any procedures.

Of the full CDaCI cohort, over 75% of participants in both groups completed this second IQ testing at one of their annual follow-up visits between ages 6–13 years old ($n = 147$ CI, $n = 75$ NH). For the CI group, 6% did not complete the assessment because they moved and 8% were lost to follow-up. In the NH, 3% moved and 14% were lost to follow-up. Among the participants with CIs, based on parent report, 11 had been diagnosed with one or more additional comorbidities, including autism spectrum disorder, cerebral palsy, CHARGE syndrome, encephalitis, hypogenesis of the corpus callosum, Noonan syndrome, periventricular leukomalacia, pervasive developmental disorder, and seizure disorder. These were considered more severe developmental disorders or medical conditions that would likely affect cognitive development. In contrast, children in the CI ($n=7$) and NH ($n=4$) groups with Attention Deficit Hyperactivity Disorder (ADHD) or other learning disorders were considered to have more mild conditions that are not commonly related to deficits in intellectual functioning. The 11 participants in the CI + Comorbidities group were examined

separately from the remaining 136 participants in the CI group who were typically developing.

Table 1 displays demographic and clinical characteristics of the CI group, the NH group, and the CI + Comorbidities group. The CI and NH groups did not statistically differ on age at study enrollment, gender, race, ethnicity, or mode of communication at the time of WISC testing. Of note, parents reported on their child's mode of communication. Therefore, some parents of children in the NH group also reported that their child was exposed to auditory-verbal and auditory-oral communication (see Table 1). The CI group was significantly older (mean 8.5 years) than the NH group (mean 7.7 years) at the time of the WISC administration and had lower maternal education and annual household income at baseline. Compared to the CI + Comorbidities group, the CI group was younger at age at onset of deafness (mean 2.5 vs 4.2 months). There were no other statistically significant differences between these groups.

Procedures

Study visits were completed in 1 or 2 days, depending on family schedules and travel burden. Families were given honoraria for each year of their participation and gift cards after each completed visit. Families in the CI group were also provided with a 2-year extension of their child's CI processor warranty after completing 3 years of follow-up. A large assessment battery was administered, including measures of receptive and expressive language, psychosocial functioning, cognitive/executive functioning, and visual attention. An IQ evaluation was conducted at baseline prior to enrollment in the study, using the Bayley Psychomotor Development Index (PDI) and Mental Development Index or the Leiter International Performance Scale-Revised. An intellectual assessment was conducted again during the school-age period (ages 6–13 years), using the Wechsler Intelligence Scale for Children, 4th Edition (WISC-IV). This enabled us to evaluate a major aim to compare possible changes in IQ over time and between tests in the same sample of children.

Measures

Bayley Scales of Infant and Toddler Development, 2nd Edition (BSID-II) (Bayley 1993)—The Bayley is a developmental assessment of children's mental and motor development for ages 1 to 42 months. The examiner presents a series of test materials to the child and observes the child's responses and behaviors. It takes 45–60 minutes to complete. This assessment evaluates children along three scales: Mental, Psychomotor Development (PDI), and Behavior. We administered the Mental and Psychomotor Development Scales; however, for our major analyses, we relied on the PDI which assesses the degree of body control, large muscle coordination, and finer manipulatory skills of the hands and fingers. The PDI has been found to be most predictive of later IQ (van Batenburg-Eddes 2013; Hitzert et al. 2014; Ghassabian 2016). The BSID are known to have high reliability and validity. The mental and motor scales have high correlation coefficients (.83 and .77 respectively) for test-retest reliability.

Leiter International Performance Scale-Revised (Leiter-R) (Roid & Miller 1997)—The Leiter-R is an individually-administered test designed to assess cognitive function in

children ages 2 years and older, adolescents, and adults up to age 20. It has been validated for individuals with hearing loss or language delays because no verbal responses are required to complete testing. The Leiter-R includes measures of nonverbal intelligence in fluid reasoning and visualization, as well as appraisals of nonverbal memory, attention, and cognitive interference. In this study, we administered the four subtests that make up the Brief IQ Composite (Figure Ground, Form Completion, Repeated Patterns, and Sequential Order). Adequate to strong correlations have been found between the Leiter BIQ and Full Scale IQ. It has demonstrated strong reliability and validity (Roid et al. 2003).

Wechsler Intelligence Scale for Children (WISC-IV) (Wechsler 2003)—Cognitive abilities were measured using the WISC-IV when children were ages 6–13. All raw scores were summed and converted into scaled scores for each subtest. Subtest scaled scores were summed and converted into standard scores for index scores. For this study, intelligence was measured using two indices: Perceptual Reasoning (PRI) and Processing Speed (PSI). The Verbal Comprehension and Working Memory indices were not included because they require oral language skills, which can penalize children with hearing loss. Definitions of classifications on the WISC are as follows: Borderline = 79; Low Average = 80–89; Average = 90–109; High Average = 110.

Comprehensive Assessment of Spoken Language (CASL) (Carrow-Woolfolk 1999)—Oral language was measured using the CASL, which was developed to assess a wide range of receptive and expressive language skills for children ages 3–22. This study utilized the core composite score, which consisted of 4 or 5 subtests, depending on the child's age. The core composite is comprised of the subtests that theoretically and developmentally best represent language skills at a given age and is designed to capture the underlying skills required for spoken language, which can be divided into 4 structural categories (Lexical/Semantic, Syntactic, Supralinguistic, and Pragmatic). Raw scores from the core subtests are summed and converted to an overall Core Composite standard scores. Definitions of classifications on the CASL are as follows: Below Average < 85; Average = 85–115; Above Average > 115.

All of the IQ and language measures used in this study are age-standardized and have a mean of 100 and standard deviation of 15 (normative data), with higher scores indicating better performance.

Statistical analyses

Baseline demographic and clinical characteristics, as well as intelligence and language scores, were presented as means and standard deviations for continuous variables and as numbers and percentages for categorical variables. For continuous variables, comparisons were conducted first between the CI and NH groups and then between the CI and CI + Comorbidities groups using either t-tests or Wilcoxon-Mann-Whitney tests. For categorical variables, separate comparisons between the CI and NH groups and the CI and CI + Comorbidities groups used Fisher's exact tests. We also examined categorizations of the CI group in relation to the normative data for the WISC and CASL. For comparisons of the CI vs. NH groups we used a Bonferroni correction procedure to guard against increasing Type I

error rate (Armstrong 2014). For comparisons of the CI vs. CI + Comorbidities groups, we only calculated differences between the composite scores (Baseline IQ, PRI, PSI, and CASL Core Composite). Linear regression was used to control for maternal education when comparing the IQ scores of the groups; in addition, hierarchical multiple regression was used to examine the contributions of hearing status and language on IQ scores. Pairwise correlations between IQ scores at both time points, demographic and clinical characteristics, and language performance were examined separately for each of the three groups. A composite IQ was used at baseline (pre-implantation), utilizing scores from the Bayley PDI or the Leiter BIQ, depending on the child's age. Stata 11 (StataCorp, College Station, TX) was used for all analyses. For all group comparisons we used a $p < 0.004$ to correct for multiple tests (i.e., Bonferroni correction); correlational analyses were evaluated using $p < 0.05$.

Results

CI vs. NH Comparisons

As shown in Table 2, the CI group scored significantly lower on the Baseline IQ compared to the NH group (101.3 vs 107.9; $p < 0.004$). Despite these lower scores, both groups scored above the normative mean of 100. Similarly, we found statistically significant differences between these groups at the school-age assessment on both the PRI and PSI indices ($p < 0.001$). However, both groups' average scores were within one standard deviation of the age-matched normative sample. On the PRI, the CI group scored 100.3 (at the normative mean), compared to the NH group's score of 115.5 (one SD above the mean). On the PSI, the CI group scored 96.7, slightly below the normative mean, while the NH group scored 103.4. After controlling for maternal education, the CI group scored 13.7 points lower than the NH group ($p < 0.001$) on the PRI and 6.3 points lower than the NH group ($p = 0.004$) on the PSI.

On the WISC subtests, the CI group's subscale scores ranged from 9.1 (Picture Completion) to 10.4 (Matrix Reasoning), while in the NH group, subtest scores ranged from 10.2 (Coding) to 13.1 (Matrix Reasoning). The CI group scored statistically significantly lower than the NH group across all subtests ($p < 0.001$), except Coding and Cancellation on the PSI. After controlling for maternal education, all the previous differences on subtests remained significantly different ($p = 0.001$).

Qualitative classifications of the WISC and CASL scores by group are displayed in Table 3. In addition, comparisons were made between the CI group and normative distributions on these measures. Approximately half of the CI children scored in the average range on PRI and PSI. In contrast, a larger proportion of the NH group scored in the high average range (73.3%) on PRI. Note that the two groups were more similar on the PSI composite. In general, children in the CI group performed similarly to the normative distribution, with the exception of Picture Completion, on which more children with CIs scored in the Borderline and Low Average range.

As expected, CASL Core Composite scores differed between the CI and NH groups (CI mean = 77.2, SD=24.4; NH mean = 115.7, SD = 18.2) as well as the normative distribution.

Children with CIs scored significantly lower ($p < 0.001$) than the NH group and the normative data. For the CI group, 59% scored in the Below Average range on the CASL in comparison to the normal hearing group (5.5% in this range).

Correlations between demographics, IQ scores, and language scores

Correlations between demographic variables, IQ scores at both test ages, and school-age language scores are shown in Table 4. Among the CI group, age at study enrollment and CI activation age were correlated with Baseline IQ but not WISC scores. Gender and income were not correlated with IQ scores at either time point. Maternal education was not correlated with Baseline IQ, but it was significantly correlated with WISC PRI and PSI. As expected, Baseline IQ was minimally correlated with school-age WISC PSI; unexpectedly, however, Baseline IQ was not correlated with WISC PRI. Both WISC composite scores were significantly correlated with CASL scores.

In the NH group, age at study enrollment and gender were not correlated with IQ scores at either time point. However, income was correlated with Baseline IQ and WISC PRI but not PSI. Maternal education was significantly correlated with the WISC PRI, but not with Baseline IQ or WISC PSI. As expected, Baseline IQ was correlated with the both the school-age WISC PRI and PSI. The WISC PRI score was significantly correlated with the CASL Core Composite score assessed at the same visit, but the WISC PSI score was not correlated with language scores.

Hierarchical Regression Models Predicting IQ

A hierarchical regression analysis was performed to examine the contributions of hearing status (CI vs NH), maternal education, and language on the PRI and PSI scores across groups. Results are summarized in Table 5. For the initial block we entered hearing status, followed by maternal education, CASL scores, and the interaction between hearing status and language scores. The well-established predictors of hearing status and maternal education on IQ explained 25.9% of the variance in PRI scores. Adding the CASL composite explained an additional 9% of the variance. Interestingly, the interaction between hearing status and language scores did not contribute to the explained variance in PRI performance. A total of 35.2% of the variance was explained by this combination of variables. Thus, language was the primary driver of the variation seen in PRI scores in both groups, with hearing status and maternal education dropping out of the final model.

A similar model was tested for PSI scores across groups. The well-established predictors of hearing status and maternal education explained only 6.4% of the variance in PSI scores. Adding the CASL composite explained an additional 8.3% of the variance. Similarly, the interaction between hearing status and language scores did not contribute to the explained variance in PSI performance. A total of 15.6% of the variance was explained by this combination of variables. Thus, language explained the largest proportion of the variation; however, note that this combination of variables accounted for only half of the total variance in PSI vs PRI.

CI vs. CI + Comorbidities Comparisons

For exploratory purposes, we examined differences between our CI and CI + Comorbidities groups on the baseline and school-age composite scores only (see Table 2). At baseline, the IQ scores differed between these groups, with the CI group scoring significantly higher than the CI + Comorbidities group. This difference remained significant after controlling for maternal education ($p = 0.001$). While school-age WISC PSI scores significantly differed between the groups, evidencing the same pattern, WISC PRI scores and CASL language scores did not statistically differ between the groups.

Discussion

Our overall objective was to evaluate differences in IQ from early childhood to the school-age period in children with CIs versus hearing peers. Significant group differences were found in the Baseline IQ between these two groups, with children with CIs scoring lower than those with typical hearing. However, as hypothesized, both groups' scores were in the average range and the group differences were likely due to the higher socioeconomic status of the normal hearing group. These results are similar to those found in previous studies comparing children with hearing loss to those with normal hearing (Khan et al. 2005; Geers et al. 2008; De Giacomo et al. 2013). The later evaluation of IQ in school-age children replicated these results, finding significant differences between groups, both scoring in the normal range. On the WISC measures, children using CIs performed similarly across subtests on the PRI and PSI.

As our second aim hypothesized, we found minimal to modest associations between IQ measured in early childhood and during the school-age period. Specifically, these associations were stronger in the hearing group. Possibly early IQ measures are highly influenced by language and stimulation provided in the home environment. This indicates that early IQ measures are not predictive of future cognitive functioning in children with hearing loss. This has also been reported by Meinzen-Derr and colleagues (2017), which reported that IQ assessments conducted prior to 24 months are not as predictive of later IQ as assessments conducted after 24 months. These early IQ measures may be used to help identify targets for early intervention, but should not be the only measure used to determine whether a child will benefit from a specific type of amplification and/or educational placement (Cruz et al. 2012). Further, because children with hearing loss are at risk for language delays, the ability to identify potential developmental delays early becomes vital to successful language intervention (Meinzen-Derr et al., 2017).

As expected, children with CIs and additional comorbidities scored significantly lower at baseline and on the WISC PSI at the follow-up assessment compared to peers with CIs not reporting additional comorbidities. Processing speed reflects the automatic processing of information, speed and fluency in tasks, and ability to concentrate and focus. Importantly, these cognitive skills have been shown to predict academic performance in reading and math (Geers et al. 2008); thus, placing these children at greater risk for falling behind in school. The CI group with versus without additional comorbidities also scored lower on the WISC PRI composite, though this difference did not reach statistical significance. Children with additional comorbidities evidenced more variability at baseline and on the WISC PRI,

suggesting that although the group means were not statistically different, a large proportion of children with additional disabilities were scoring far below the mean. Surprisingly, this does not appear to be mediated by language differences on the CASL. These findings indicated that, although these children evidenced minimal cognitive deficits during early childhood, specific developmental delays emerged later and were diagnosed after implantation.

Consistent with prior literature we found modest, positive associations between maternal education and school-age IQ scores. Mothers with higher education had children who scored better on perceptual reasoning tasks. In the CI group, these associations were found for both Perceptual Reasoning and Processing Speed indices. However, in the hearing group no association was found between this variable and processing speed. One possible reason for these contrasting results is that the Perceptual Reasoning Index, which focuses on logic, utilizing fluid reasoning and solving novel problems relies on stimulation and input from the child's home environment. Mothers with less education who have children with hearing loss have been shown to provide less cognitive and linguistic stimulation (Cruz et al. 2013; Quittner et al. 2013). These results support the need for early intervention programs that focus on the quality of input in the parent-child dyads (Cejas & Quittner in press). We have recently developed a parent training intervention to improve the quality of parent-child relationships and use of higher-level cognitive and linguistic strategies (Parent-Child Early Approaches to Raising Language Skills; PEARLS). This is particularly important for families coming from a lower socioeconomic background.

Evaluation of school-age language scores revealed statistically significant differences between the CI and hearing group; as expected children with CIs had substantially lower mean scores on the CASL when compared to those with normal hearing. In contrast, no statistically significant differences were found between the two CI groups. Both of these groups scored within the impaired range. Importantly, even though children with CIs who had additional comorbidities had relatively severe conditions (e.g., Autism, Cerebral Palsy), they demonstrated benefits in terms of their oral language development (Cruz et al. 2012). Despite these benefits, they continue to perform below age-related norms. These results are clinically important given that over half of children receiving cochlear implants to date have additional comorbidities.

As is well-documented in the language literature, children from families from lower socioeconomic status evidence worse performance on standardized language assessments, and this also applies to children with hearing loss (Niparko et al. 2010). In our study, we found moderate to strong associations between maternal education and language scores for both the CI and hearing groups. Furthermore, relationships between language and Baseline IQ were only found for the normal hearing group. In contrast, significant, moderate associations were found between school-age IQ scores and language on both indices for the CI group. However, for the hearing group only the PRI composite was correlated with language scores. This suggests that these IQ measures in children with hearing loss are confounded by language performance (Geers et al. 2007; Kammerer et al. 2010; Reesman et al. 2014). The regression models also inform this discussion because they evaluated the relative contribution of hearing status, maternal education, and language on school-age IQ.

Importantly, hearing status and maternal education were no longer predictors of IQ once spoken language performance was taken into account. This reveals the importance of early interventions that boost language and communication (e.g., oral or sign). Families from economically disadvantaged backgrounds are at-risk for worse outcomes, but can clearly benefit from early intervention programs that emphasize language and parent training (Cejas et al. in press).

These results support the theory that although nonverbal assessments minimize the impact of auditory deficits and eliminate the need for verbal instructions, they tap into the global factor of intelligence. This relates to the constructivist approaches that expect that language-learning processes show commonalities with nonlinguistic learning (Deak, 2014). Thus, it appears that variability in language may be related to variability in cognitive skills. Specifically, we hypothesize that nonverbal IQ is related to language via working memory. Nonverbal measures focus on visual reasoning, visual discrimination, and abstract visual problem-solving, which is impacted by working memory that affects comprehension, production, and learning (Deak, 2014; Phillips et al., 2014).

Limitations

Although this is the largest longitudinal and diverse study comparing children with CIs and normal hearing on their intelligence and language performance, it had several limitations. First, our normal hearing comparison group included families with higher socioeconomic status, leading to their higher scores on both the IQ and language measures. This difference in part explains the difference found in school-age IQ scores between the CI and normal hearing groups, despite the CI children performing in the average range. Despite the interesting cognitive differences we found between the CI and CI + Comorbidities groups, our sample size was small. In addition, the comorbidity group was composed of children with more severe conditions, such as autism, which emphasized their deficits. Children with milder conditions, such as ADHD, were included in both groups. In general, children with higher IQ performed better on language assessments. Lastly, although we utilized the nonverbal indices of the WISC, which are frequently used to evaluate intelligence in children with hearing loss, we were surprised by the substantial associations with language. These associations indicate that although the WISC PRI and PSI are considered “nonverbal” measures of intelligence, the instructions and administration are verbally mediated which can lead to misunderstandings, confusion, and lower performance in children with hearing loss (Kammerer et al. 2010; Reesman et al. 2014).

Recommendations for Use of Nonverbal Intelligence Tests in Children with Hearing Loss

Assessing intelligence in children with hearing loss is difficult and has several unique challenges. These include evaluating the link between language skills and intelligence, adapting the assessment to meet the needs of the child (e.g., administration for children with severe language delays or other communication modes), and appropriate interpretation of the results. As mentioned above, most intelligence tests rely heavily on verbal communication for instructions and responses (Reesman et al. 2014). For example, although the WISC Perceptual Reasoning and Processing Speed indices use nonverbal stimuli, the processing and comprehension of the instructions require verbal skills. To ensure the child's

understanding, the clinician may alter the test administration. However, such deviations can make the test results unreliable. In addition, underestimating a child's IQ can have real world consequences for his or her classroom placement or accommodations. Importantly, the majority of children with hearing loss have normal intelligence and can be effectively accommodated in a mainstream classroom or deaf or hard of hearing program.

Thus, we recommend use of purely nonverbal intelligence tests, such as the Leiter International Performance Scale, that were specifically designed for children with hearing loss or language delays, particularly when assessing for educational eligibility. Although previous research has shown that verbal intelligence translates better to academic achievement in children with hearing loss (Wood & Dockrell 2010), we would recommend the use of functional assessments that are more closely related to academic skills (e.g., Woodcock Johnson Tests of Achievement, Wechsler Individual Achievement Test).

In addition to administrative challenges, few intelligence tests have normative data based on children with hearing loss (Reesman et al. 2014). For example, the commonly used WISC does not have normative data for children with hearing loss, and thus, their scores are compared to normal hearing children. Use of these norms with hearing-impaired children disadvantages them in relation to interpretation and subsequent clinical recommendations. Moreover, it is also important not to set expectations too low, which could affect goal setting at home or at school. However, given that a large proportion of children with cochlear implants are in mainstream settings, these assessments may provide an estimation of how the child functions in these settings.

In sum, it is imperative to recognize that intellectual assessments of children who are deaf should be individualized. All assessments should be adapted based on the nature of the referral question, background of the child, goals of the evaluation, and language skills and fluency of the child (Reesman et al. 2014). Although early developmental assessments are helpful for understanding a child's strengths and needs to guide treatment planning, it is critical to monitor the child's progress over time. Meinzen-Derr and colleagues (2017) reported that assessments completed at 24 months of age or later are better predictors of later IQ than assessments administered at earlier ages. In addition, some developmental disabilities are not evident in infancy or toddlerhood and thus, these early developmental assessments may not reliably identify all children who will go on to have a diagnosis.

The major conclusion from our study is that children using cochlear implants evidenced normal IQ on an intelligence measure that is language dependent. However, we strongly recommend the use of nonverbal intelligence tests that are designed and normed for children with hearing loss and/or language delays. At-risk groups, including families from lower socioeconomic backgrounds and children with additional comorbidities, could clearly benefit from early implantation and parent-based early interventions (Cruz et al. 2012; Quittner et al. 2013; Cejas et al. in press).

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Table 1

Demographics and clinical characteristics of the CDaCI subsample

Characteristic	CI (n = 136)	NH (n = 75)	CI + Comorbidities (n = 11)	CI vs. NH	CI vs. CI + Comorbidities
Age of onset (months), mean (SD)	2.5 (7.0)	-	4.2 (5.5)	-	0.03*
Age at diagnosis (months), mean (SD)	10.0 (10.0)	-	12.0 (7.8)	-	0.36
Length of hearing aid use before implant (months), mean (SD)	15.0 (12.9)	-	13.5 (13.3)	-	0.72
Pure tone average in better ear – unaided, mean (SD)	104.7 (16.2)	15.5 (6.3)	103.8 (20.2)	<0.001**	0.90
Pure tone average in better ear – aided, mean (SD)	73.8 (22.3)	-	74.3 (25.8)	-	0.70
Age at study enrollment (months), mean (SD)	2.2 (1.2)	2.3 (1.0)	2.2 (1.2)	0.53	0.89
Age at implant activation (years), mean (SD)	2.4 (1.2)	-	2.4 (1.1)	-	0.86
Age at WISC administration (years), mean (SD)	8.5 (1.5)	7.7 (0.7)	8.8 (1.8)	<0.001**	0.67
Onset of hearing loss, n (%)					0.07
Sudden	7 (5.2)	-	2 (18.2)		
Progressive	45 (33.1)	-	5 (45.5)		
Congenital	78 (57.4)	-	3 (27.3)		
Other (e.g. unknown)	6 (4.4)	-	1 (9.1)		
Mode of Communication: Home (at time of WISC testing), n (%)				0.03*	0.99
Sign communicator	1 (0.8)	0 (0)	0 (0)		
Speech and sign communicator	18 (13.5)	2 (3.1)	1 (9.1)		
Speech communicator	114 (85.7)	63 (96.9)	10 (90.9)		
Mode of Communication: School (at time of WISC testing), n (%)				0.12	0.50
Simultaneous - sign emphasis	6 (4.7)	0 (0)	0 (0)		
Simultaneous - equal emphasis	7 (5.5)	0 (0)	0 (0)		
Simultaneous - speech emphasis	12 (9.4)	2 (4.1)	1 (9.1)		
Cued speech	1 (0.8)	1 (2.0)	0 (0)		
Auditory-oral	35 (27.3)	12 (24.5)	6 (54.6)		
Auditory-verbal	67 (52.3)	34 (69.4)	4 (36.4)		
Gender, n (%)				0.89	0.54
Male	60 (44.1)	34 (45.3)	6 (54.6)		
Female	76 (55.9)	41 (54.7)	5 (45.5)		
Race, n (%)				0.55	0.76

Characteristic	CI (n = 136)	NH (n = 75)	CI + Comorbidities (n = 11)	CI vs. NH	CI vs. CI + Comorbidities
White	105 (77.2)	59 (78.7)	9 (81.8)		
African American	11 (8.1)	9 (12.0)	0 (0)		
Asian	6 (4.4)	1 (1.3)	1 (9.1)		
Other	14 (10.3)	6 (8.0)	1 (9.1)		
Ethnicity, n (%)				0.18	0.22
Hispanic	27 (19.9)	9 (12.0)	0 (0)		
Non-Hispanic	109 (80.2)	66 (88.0)	11 (100)		
Maternal education, n (%)				0.01 *	0.43
< High school	8 (5.9)	3 (4.0)	0 (0)		
High school graduate	20 (14.7)	2 (2.7)	0 (0)		
Some college or college graduate	108 (79.4)	70 (93.3)	11 (100)		
Annual household income, n (%)				<0.001 **	0.52
\$15,000	11 (8.1)	3 (4.0)	0 (0)		
\$15,000 – 29,999	16 (11.8)	2 (2.7)	2 (18.2)		
\$30,000 – 49,999	28 (20.6)	4 (5.3)	2 (18.2)		
\$50,000 – 74,999	25 (18.4)	14 (18.7)	1 (9.1)		
\$75,000 – 100,000	17 (12.5)	10 (13.3)	4 (36.4)		
> \$100,000	23 (16.9)	36 (48.0)	1 (9.1)		
Don't know/declined	16 (11.8)	6 (8.0)	1 (9.1)		

* p < 0.05,

** p < 0.01

Table 2

Means and standard deviations of IQ and language scores by group

Test	CI (n = 136)	NH (n = 75)	CI + Comorbidities (n = 11)	CI vs. NH	CI vs. NH adjusted [^]	CI vs. CI + Comorbidities	CI vs. ND
Bayley MDI	83.7 (13.8) [n = 93]	106.5 (10.0) [n = 34]	72.8 (19.0) [n = 9]	<0.001*	<0.001*	<0.001*	<0.001*
Bayley PDI	98.9 (16.2) [n = 93]	106.7 (16.0) [n = 35]	76.2 (22.8) [n = 9]	0.009	0.02		0.27
Leiter Brief IQ	106.8 (18.1) [n = 41]	108.9 (10.4) [n = 40]	111.0 (11.3) [n = 2]	0.26	0.48		0.01
Baseline IQ ⁺	101.3 (17.1)	107.9 (13.3)	82.5 (25.0)	0.002*	0.004	<0.001*	0.18
WISC PRI	100.3 (15.3)	115.5 (13.3)	93.3 (24.3)	<0.001*	<0.001*	0.08	0.40
Block Design	9.9 (2.8)	11.6 (2.8)		<0.001*	<0.001*		0.39
Picture Concepts	9.8 (3.4)	12.8 (2.8)		<0.001*	<0.001*		0.23
Matrix Reasoning	10.4 (3.1)	13.1 (2.8)		<0.001*	<0.001*		0.09
Picture Completion	9.1 (2.4)	10.9 (2.3)		<0.001*	<0.001*		<0.001*
WISC PSI	96.7 (15.0)	103.4 (14.3)	83.3 (13.5)	<0.001*	0.004	0.002*	0.006
Coding	9.3 (3.4)	10.2 (3.5)		0.04	0.12		0.01
Symbol Search	9.4 (3.3)	11.0 (2.7)		<0.001*	0.001*		0.03
Cancellation	9.6 (3.0)	10.3 (2.9)		0.048	0.11		0.05
CASL Core Composite	77.2 (24.4)	115.7 (18.2)	71.9 (24.4)	<0.001*	<0.001*	0.26	<0.001*

ND = normative data, where mean = 100, SD = 15 or mean = 10, SD = 3 for WISC subscales

* $p < 0.004$ (statistically significant after Bonferroni correction)

[^] After controlling for maternal education

⁺ Combines participants tested with Bayley PDI & those tested with Leiter

Table 3Wechsler and CASL qualitative classifications by group, *n* (%)

	CI (<i>n</i> = 136)	NH (<i>n</i> = 75)	CI vs. NH	CI vs. ND ⁺
WISC Perceptual Reasoning Index (PRI)			<0.001*	0.94
Borderline (79)	11 (8.2)	1 (1.3)		
Low Average (80–89)	24 (17.8)	0 (0)		
Average (90–109)	63 (46.7)	19 (25.3)		
High Average (110)	37 (27.4)	55 (73.3)		
Block Design			0.003*	0.59
Borderline (5)	7 (5.2)	1 (1.3)		
Low Average (6–7)	20 (14.7)	6 (8.0)		
Average (8–11)	69 (50.7)	27 (36.0)		
High Average (12)	40 (29.4)	41 (54.7)		
Picture Concepts			<0.001*	0.52
Borderline (5)	12 (8.9)	1 (1.3)		
Low Average (6–7)	20 (14.8)	3 (4.0)		
Average (8–11)	58 (43.0)	14 (18.7)		
High Average (12)	45 (33.3)	57 (76.0)		
Matrix Reasoning			<0.001*	0.25
Borderline (5)	5 (3.7)	0 (0)		
Low Average (6–7)	19 (14.1)	1 (1.3)		
Average (8–11)	68 (50.4)	22 (29.3)		
High Average (12)	43 (31.9)	52 (69.3)		
Picture Completion			0.001*	0.045
Borderline (5)	6 (4.7)	0 (0)		
Low Average (6–7)	28 (21.9)	4 (8.2)		
Average (8–11)	76 (59.4)	26 (53.1)		
High Average (12)	18 (14.1)	19 (38.8)		
WISC Processing Speed Index (PSI)			0.006	0.24
Borderline (79)	17 (12.8)	1 (1.3)		
Low Average (80–89)	22 (16.5)	12 (16.0)		
Average (90–109)	73 (54.9)	40 (53.3)		
High Average (110)	21 (15.8)	22 (29.3)		
Coding			0.20	0.62
Borderline (5)	16 (11.9)	6 (8.1)		
Low Average (6–7)	21 (15.7)	11 (14.9)		
Average (8–11)	71 (53.0)	33 (44.6)		
High Average (12)	26 (19.4)	24 (32.4)		
Symbol Search			0.005	0.59
Borderline (5)	18 (13.4)	2 (2.7)		
Low Average (6–7)	17 (12.7)	3 (4.0)		
Average (8–11)	64 (47.8)	42 (56.0)		

	CI (n = 136)	NH (n = 75)	CI vs. NH	CI vs. ND ⁺
High Average (12)	35 (26.1)	28 (37.3)		
Cancellation			0.34	0.78
Borderline (5)	10 (7.9)	3 (4.3)		
Low Average (6–7)	17 (13.4)	8 (11.4)		
Average (8–11)	71 (55.9)	35 (50.0)		
High Average (12)	29 (22.8)	24 (34.3)		
CASL Core Composite			<0.001*	<0.001*
Below Average (< 85)	78 (58.7)	4 (5.5)		
Average (85–115)	46 (34.6)	29 (39.7)		
Above Average (> 115)	9 (6.8)	40 (54.8)		

⁺ND=normative distribution; WISC normative distribution: Borderline = 9.1%, Low Average = 16.1%, Average = 49.5%, High Average = 25.3%; CASL normative distribution: Below Average = 15.9%, Average = 68.3%, Above Average = 15.9%

Table 5

Hierarchical regression models predicting IQ

	Perceptual Reasoning Index				Processing Speed Index			
	β	t	R ²	R ²	β	t	R ²	R ²
Block 1			0.20	0.20			0.05	0.05
Hearing status (NH vs CI)	15.16	7.21 ^{***}			6.69	3.15 ^{**}		
Block 2			0.26	0.06			0.06	0.02
Hearing status (NH vs CI)	13.67	6.6 ^{***}			6.30	2.92 ^{**}		
Maternal edu: hs grad vs not	1.94	0.4			7.15	1.32		
Maternal edu: any college vs < hs	12.24	2.8 ^{**}			8.87	1.94		
Block 3			0.35	0.09			0.15	0.08
Hearing status (NH vs CI)	5.55	2.17 [*]			-1.00	-0.37		
Maternal edu: hs grad vs < hs	-2.99	-0.58			5.98	1.11		
Maternal edu: any college vs < hs	3.53	0.76			5.00	1.04		
Language standard score (1 pt increase)	0.24	5.25 ^{***}			0.19	4.09 ^{***}		
Block 4			0.35	0.0001			0.16	0.009
Hearing status (NH vs CI)	3.32	0.3			15.10	1.34		
Maternal edu: hs grad vs < hs	-2.98	-0.57			5.89	1.09		
Maternal edu: any college vs < hs	3.50	0.75			5.16	1.07		
Language standard score (1 pt increase)	0.23	4.65 ^{***}			0.23	4.33 ^{***}		
Hearing status × language score (NH, 1 pt increase)	0.02	0.21			-0.15	-1.47		

* $p < 0.05$,

** $p < 0.01$,

*** $p < 0.001$