AMERICAN ACADEMY OF OTOLARYNGOLOGY–HEAD AND NECK SURGERY FOUNDATION

Otolaryngology–Head and Neck Surgery
2021, Vol. 165(5) 731–738

DOI: 10.1177/0194599821989642
http://otojournal.org
SAGE

Cochlear Implants in Neurologically Impaired Children: A Survey of Health-Related Quality of Life

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Abstract

Objectives. Evaluate health-related quality-of-life (HRQoL) measures in noncommunicative, neurologically impaired, developmentally delayed (NCNIDD) children compared to normally developing children (ND) who undergo cochlear implantation (CI).

Study Design. Cross-sectional survey of parents of NCNIDD and ND children who underwent CI.

Setting. Two tertiary care medical centers.

Methods. Questions comprising the Children With Cochlear Implants: Parental Perspectives survey were used in analysis. Average responses were calculated within 8 domains (communication, general functioning, self-reliance, well-being and happiness, social relationships, education, effects of implantation, and support the child). Groups were compared using Wilcoxon rank-sum test. Impact of individual and collective socioeconomic/family covariates was assessed using analysis of variance.

Results. Surveys were returned from 17 of 42 (40%) patients with NCNIDD and 35 of 131 (27%) patients with ND. There were no statistically significant differences between groups in survey response rate, age, sex, age at implantation, current age, or duration of implant use. Overall, parents of children with ND responded more favorably in all domains vs children with NCNIDD. Parents of children with NCNIDD answered neutrally or favorably in all domains, except "support the child" and "self-reliance" domains. Differences between groups in mean domain scores, univariably and almost universally when adjusting for socioeconomic and family variables individually and collectively, were statistically significant.

Conclusions. This study suggests that HRQoL benefits of CI are perceived in most domains by parents of children with NCNIDD, albeit less strongly than children with ND. A survey sensitive to challenges of children with NCNIDD may better capture benefits that may not be apparent in this study.

Keywords

health-related quality of life, neurologically impaired, cochlear implantation

Received September 25, 2020; accepted January 3, 2021.

In most healthy children, there is robust evidence for benefit in speech and language development with cochlear implantation (CI).[1-3] However, determining auditory or speech and language outcomes is difficult in children who are noncommunicative, neurologically impaired, and developmentally delayed (NCNIDD). These children do not have the cognitive ability to participate in the testing or the ability to reliably communicate their perceptions. The outcomes of these patients are largely focused on sound awareness and safety concerns, and decision for implantation is based at least partially on parental rationale to improve quality of life in the children. Quantifying the quality-of-life benefits of CI in these children would allow for validation of these subjective and anecdotal improvements seen by families.

Health-related quality-of-life (HRQoL) assessments focus on an individual's perception of physical health, mental health, and social well-being. There are validated HRQoL questionnaires for pediatric populations related to hearing loss[4] and specific surveys that evaluate CI-related QoL, but these are limited for use in adults.[5] Condition-specific issues for CI exist that are not pertinent to hearing loss, including the process of cochlear implantation and the impact of the device itself. Therefore, there is a need for a survey that assesses condition-specific CI HRQoL in children.

A systematic review of HRQoL after cochlear implantation in 2006 found only 10 studies evaluating children younger than 18 years,[6] with only 1 study that specifically surveyed children directly.[6] Since then, there have been identified 7 additional studies that evaluated children's responses directly[7] and have found similar positive

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responses to QoL.[8] The inherent difficulty in eliciting responses in young patients is reflected in the very few number of studies that have been published evaluating HRQoL by directly surveying pediatric patients with cochlear implants. Because of the difficulty in assessing quality of life in pediatric patients and furthermore in patients with NCNIDD, parents are often used as a proxy for the children. Even with parental proxy, assessing HRQoL can be just as difficult as evaluating auditory, speech, and language outcomes in these children with NCNIDD due to challenge of eliciting responses for the survey. Supplemental Table S1 (in the online version of the article) summarizes studies that have evaluated HRQoL using parents as a proxy in CI patients.[4,9-19]

With the goal of developing a CI-specific HRQoL questionnaire specific to children, Archbold et al[20] first evaluated 30 patients with an open-ended questionnaire survey of parents of children who underwent CI to derive the Parents Views and Experiences With Pediatric CI Questionnaire (PVEIQ).[21] Since then, the short form of the survey has been created and validated as the Children With Cochlear Implants: Parental Perspectives survey (see Suppl. Figure S1 in the online version of the article).[18,21,22] More recent studies have relied on this short form survey to evaluate HRQoL of healthy children with CI and have shown quality-of-life benefits (summarized in Suppl. Table S2 in the online version of the article).[4,9,22-25]

However, none of the studies evaluated children with NCNIDD. The goal of this study is to evaluate the HRQoL measures specifically in children with NCNIDD using the Children With Cochlear Implants: Parental Perspectives survey and compare them with normally developing children. The hypothesis is that although the audiologic, speech, and language outcomes may differ between the groups, parents of children with NCNIDD will perceive positive HRQoL benefits comparable to normally developing children.

Methods

Study Population

The study was conducted following approval from the Institutional Review Boards (IRBs) of the University of Pittsburgh and Cleveland Clinic Foundation. A retrospective review was completed for the years 2010 to 2020 to identify patients who have had cochlear implantation prior to age 18 years and had been implanted for a minimum of 4 months from 2 tertiary care academic hospital systems. At both institutions, activation of implant occurs within the first month after surgery. This allowed for 3 months of minimum CI use prior to the survey to allow for acclimatization. All patients who underwent cochlear implantation were identified with *Current Procedural Terminology* (*CPT*) code 69930.

Inclusion criteria for the NCNIDD group is that patients must have formal assessment of developmental delay by a neurologist or developmental pediatrician. Of these patients, only patients who are noncommunicative verbally and have neurologic impairment with limited to no cognitive ability were included. Cognition is defined as mental processes involved in gaining knowledge and comprehension. Inclusion criteria for normally developing children are children who underwent CI for hearing loss but have no additional disabilities or comorbidities related to cognition and no chronic major illnesses requiring routine hospitalizations and repeat inpatient treatments (such as patients with cancer).

Surveys were initially dispensed through email solicitation. If there was no response, an email reminder was completed. Within the NCNIDD cohort, because of smaller cohort size, if there was still no response, the patient was called by one of the study team members using an IRB approved phone script and asked to participate in the study. A link was provided through email, and the questionnaire was distributed via REDCap database. Reminders were issued to any parent who did not return the questionnaire within 2 weeks of its date of issue and within 4 weeks of initial issue. Last, when there was no response, a paper copy with a postmarked envelope for return was mailed to families.

Aside from survey results, the patient's current age and duration of implant use in years were noted. Last, demographic and family-related variables were collected, including race, number of children in the house, and median household income and education based on the ZIP code of family's address using census data.

Questionnaire Used

PVEIQ, a validated 107-question closed-format questionnaire assessing parental views following cochlear implantation of their child, was initially used for data gathering. Of these, the 74 questions that comprise the Children With Cochlear Implants: Parental Perspectives survey were used in analysis.[14] Parents answered questions that were divided into 10 domains. The 10 domains were further divided into 2 main categories assessing the process of CI (process of implantation and decision to implant) and 8 measures of outcomes after CI (communication, general functioning, self-reliance, well-being and happiness, social relationships, education, effects of implantation, and supporting the child). Data covered by 34 of the questions address the process of CI (before implantation) and will not be addressed in this study. The remaining 40 of the questions can be analyzed quantitatively in these subscales addressing outcomes after CI and will be addressed as previously described in the literature.[10,23,24]

Statistical Analysis

For each individual statement, the data were summarized by the frequency of the responses and quantitative summaries using the numeric assignments in the range of 1 to 5. For positively phrased questions, a value of 1 was assigned to responses of ''strongly agree'' and the value of 5 was assigned to responses of ''strongly disagree.'' For negatively phrased questions, a value of 1 was assigned to responses of

''strongly disagree,'' while the value of 5 was assigned to responses of ''strongly agree.'' Thus, higher scores reflected a tendency toward more positive views regarding the question being asked about the CI.

Using the numeric assignments, the average response was calculated among questions within each domain for each patient. The patient groups were compared with respect to mean domain scores using the Wilcoxon rank-sum test. When a patient had missing data for a statement that was used to calculate a domain score, the domain score was then calculated based on all the remaining relevant statements that were not missing. The analysis reflects similar methodology used in previous studies that used the survey.[10,23,24] The patient groups were also compared with respect to categorical and quantitative demographic characteristics using Fisher exact tests and Wilcoxon rank-sum tests, respectively.

Age-matched analyses in 1:1 and 2:1 ratios were considered using R version 3.6.0[26] but not performed due to lack of difference with age between 2 groups and the near 2:1 control/NCNIDD cohort ratio. Analysis of impact of socioeconomic and family variables (race, median household income, median household education with percentage above high school, number of children in the house) on domain scores was completed. Last, patient's current age and duration of implant use in years and impact on domain scores were also evaluated.

The impact of individual covariates on each domain in normally developing and NCNIDD groups was assessed. Associations between 2-group forms of potential covariates and domain scores were assessed using Wilcoxon rank-sum tests. The impact of the potential covariates and the relationship between development group and mean domain scores was assessed for each domain score using analysis of variance (ANOVA) models. One model was constructed with no covariates and a separate model with each covariate individually and all together. The covariate-adjusted difference in mean domain scores between the development groups and the corresponding 95% confidence interval and *P* value were calculated based on each model.

Results

Demographics

There were 42 patients with NCNIDD identified. Surveys were returned from 17 patients (40%). Out of 132 normally developing patients who received surveys, 35 (27%) returned surveys. There was no statistically significant difference between the 2 groups in survey return rate ($P = .09$). Of the 52 parents who returned surveys, 5 were adoptive parents, and the rest were biologic parents. All surveys returned were fully completed, with only 1 survey missing mostly demographic information. Domain scores were able to be calculated for all patients.

In the normally developing group, there were 18 males and 16 females (sex not marked for 1 patient). In the patients with NCNIDD, there were 8 males and 9 females. Average age of implantation was 3.53 years for normally developing patients and 3.71 years for patients with NCNIDD. Duration of implant use was also similar, with an average of 4.86 years in normally developing and 5.50 years in the NCNIDD group. There was no statistical difference between the normally developing and NCNIDD groups in sex, age, household variables (except number of children in the house), number of patients who retained the implant, or the duration of implant use (**Table 1**). The number of children in the household was higher in the NCNIDD group (2.24 ± 0.97 vs 2.88 ± 0.86, $P = .03$). In

Table 1. Demographics and Analysis of All Returned Surveys.[a]

Group	Overall (N = 52)	Normally developing (n = 35; 67.3%)	NCNIDD (n = 17; 32.7%)	*P* value
Sex				
Male	26 (51.0)	18 (52.9)	8 (47.1)	.69
Female	25 (49.0)	16 (47.1)	9 (52.9)	
Race				
White	**40 (78.4)**	**31 (91.2)**	**9 (52.9)**	**.006**
African American	**6 (11.8)**	**2 (5.9)**	**4 (23.5)**	
Other	**5 (9.8)**	**1 (2.9)**	**4 (23.5)**	
Household variables				
Median household income	56,534.32 ± 17,969.22	56,937.10 ± 19,474.23	55,074.25 ± 11,853.25	.71
Median household education (% above high school)	91.59 ± 5.80	91.17 ± 6.34	93.10 ± 3.00	.51
Number of children in the house	**2.46±0.97**	**2.24±0.97**	**2.88±0.86**	**.029**
Age at implantation	3.59 ± 3.61	3.53 ± 3.54	3.71 ± 3.87	.82
Current age	8.69 ± 3.98	8.47 ± 3.59	9.12 ± 4.74	.77
Still have implant	50 (98)	34 (100)	16 (94)	.33
Duration of implant use, y	5.00 ± 2.48	4.86 ± 2.28	5.50 ± 3.25	.97

Abbreviation: NCNIDD, noncommunicative, neurologically impaired, and developmentally delayed.
[a]Values are presented as number (%) or mean ± SD. Values in bold reached significance.

Table 2. Overall Domain Scores.[a]

Overall domain	Overall (N = 52), mean ± SD	Normally developing (n = 35; 67.3%), mean ± SD	NCNIDD (n = 17; 32.7%), mean ± SD	*P* value
Communication	3.87 ± 0.96	4.22 ± 0.57	3.14 ± 1.18	.001
General functioning	3.61 ± 0.72	3.88 ± 0.43	3.04 ± 0.87	.001
Self-reliance	3.44 ± 0.61	3.68 ± 0.46	2.90 ± 0.58	<.001
Well-being and happiness	3.82 ± 0.58	3.97 ± 0.47	3.48 ± 0.66	.003
Social relations	3.76 ± 0.63	3.97 ± 0.47	3.34 ± 0.73	.002
Education	3.89 ± 0.72	4.17 ± 0.53	3.31 ± 0.73	<.001
Effects of implantation	3.40 ± 0.61	3.55 ± 0.59	3.09 ± 0.53	.012
Supporting the child	3.08 ± 0.61	3.27 ± 0.47	2.68 ± 0.68	.001

Abbreviation: NCNIDD, noncommunicative, neurologically impaired, and developmentally delayed.
[a]In these results, domain scores have been averaged over individual statements so that they have values from 1 to 5, with 1 representing the least favorable and 5 the most favorable responses; comparisons are performed with the Wilcoxon rank-sum test.

terms of race, there was higher percentage of white families in normally developing children in comparison to NCNIDD cohorts (91.2% and 52.9% respectively, $P = .006$).

Overall Domain Scores

When examining overall domain scores, compared to children with NCNIDD, parents of normally developing children responded with overall more favorable responses in all domains (**Table 2**). Parents of children with NCNIDD responded negatively in ''support the child'' and ''self-reliance'' domains (average scores 2.68 and 2.90 respectively). The remaining domains were on average answered neutrally or slightly favorably, with average scores ranging from 3.04 to 3.48.

Impact of Socioeconomic and Family Variables

Estimated differences in mean domain scores between groups, when adjusting for socioeconomic and family variables either individually or as a collection of covariates, were of similar magnitude to the unadjusted differences (**Table 3**). Statistical significance of the group comparisons was retained almost universally, except in ''support the child'' and ''well-being and happiness'' domains. Domains with unadjusted group differences in means of magnitude greater than 0.60 points saw significant group differences under all covariate adjustments as well. The ''support the child'' and ''well-being and happiness'' domains had a loss in statistical significance ($P = .06$) accompanied by reduced magnitude in the group difference upon covariate adjustment (from a 0.59- to 0.41-point difference and 0.50- to 0.47-point difference, respectively, when adjusting for the collection of covariates; **Table 3**). There was also loss of significance in ''effects of implantation'' when adjusted for education and duration of implant use.

Discussion

Clinical evaluation of patients who underwent cochlear implantation with speech and language measures and auditory measures have shown clear and consistent benefit.[1-3] However, the downstream effects of implantation on performance at home and social situations are not clearly understood,[4] and there are only a few studies on quality-of-life measures of the patient or the parents' perception of effects of implantation on physical and mental health (**Table 1** and **Table 2**).

In children with NCNIDD, there is significant limitation on assessing clinical capabilities with auditory, speech, and language outcomes and understanding the effects of implantation beyond clinical testing. Historically, the outcomes of these patients are largely measured with sound detection and awareness, and implantation is based on parental rationale to improve patient safety and quality of life in the children. Understanding the QoL benefits of CI in these children would allow for validation of these subjective and anecdotal improvements seen by families.

There are intrinsic difficulties in assessing HRQoL in all children. This is even more difficult in children with NCNIDD who do not have the cognitive and/or physical ability to participate in the testing and are unable to communicate their perceptions. In the study done by O'Neill et al,[21] in 2004, the authors evaluated the PVEIQ survey and felt that there was high test-retest reliability and the repeatability was satisfactory. However, the short form of the Children With Cochlear Implants: Parental Perspectives survey[21] was assessed further for validity and reliability.[18] The authors found that the α reliability of the 11 questionnaire scales varied between .41 and .74, suggesting increasing the breadth of survey may improve the α reliability but would burden families further with a longer survey. They compared parents' responses in an interview to the survey responses for content validity and found there was agreement. Last, the authors also evaluated the criterion validity by identifying contrasting cases (opposite scoring in each scale) and analyzing the descriptions obtained in the interviews. The authors found that 9 of 11 scales had support for validity.[18] Therefore, it was decided to limit analysis to the short form questionnaire since it has undergone more rigorous assessment of validity and reliability. In addition,

Table 3. Estimated Differences in Mean Domain Scores Based on Analysis of Variance Models With Covariates.[a]

Covariate	Estimated difference in mean domain score (95% CI)							
	Social relations	Education	Effects of implantation	Support the child	Communication	General functioning	Self-reliance	Well-being, happiness
No covariate	−0.63 (−0.97 to −0.30)	−0.86 (−1.22 to −0.51)	−0.46 (−0.80 to −0.11)	−0.59 (−0.92 to −0.27)	−1.09 (−1.57 to −0.60)	−0.84 (−1.20 to −0.48)	−0.78 (−1.09 to −0.47)	−0.50 (−0.82 to −0.17)
P value	.002	<.001	.012	.001	.001	.001	<.001	.003
All covariates	−0.78 (−1.22 to −0.34)	−0.80 (−1.25 to −0.34)	−0.55 (−1.04 to −0.07)	**−0.41 (−0.84 to 0.02)**	−0.84 (−1.36 to −0.32)	−0.73 (−1.26 to −0.21)	−0.90 (−1.35 to −0.45)	**−0.47 (−0.97 to 0.02)**
P value	.001	.001	.026	**.06**	.002	.008	<.001	**.06**
Sex	−0.62 (−0.96 to −0.28)	−0.86 (−1.21 to −0.51)	−0.44 (−0.76 to −0.12)	−0.61 (−0.94 to −0.29)	−1.08 (−1.57 to −0.60)	−0.84 (−1.20 to −0.48)	−0.78 (−1.09 to −0.47)	−0.49 (−0.82 to −0.17)
P value	.001	<.001	.008	<.001	<.001	<.001	<.001	.004
Race (white vs nonwhite)	−0.68 (−1.05 to −0.30)	−0.85 (−1.25 to −0.45)	−0.48 (−0.85 to −0.10)	−0.51 (−0.88 to −0.15)	−1.06 (−1.61 to −0.51)	−0.77 (−1.18 to −0.36)	−0.77 (−1.11 to −0.43)	−0.44 (−0.79 to −0.08)
P value	.001	<.001	.013	.007	<.001	<.001	<.001	.018
Median household income for ZIP code ≥$55,000	−0.82 (−1.23 to −0.41)	−0.80 (−1.25 to −0.34)	−0.48 (−0.96 to 0.00)	−0.50 (−0.96 to −0.05)	−0.88 (−1.52 to −0.23)	−0.78 (−1.28 to −0.27)	−0.94 (−1.37 to −0.51)	−0.55 (−1.02 to −0.07)
P value	<.001	.001	.05	.032	<.009	<.003	<.001	.026
Education for ZIP code (educational attainment: % high school graduate or higher) ≥92%	−0.79 (−1.21 to −0.36)	−0.79 (−1.23 to −0.34)	**−0.46 (−0.94 to 0.02)**	−0.49 (−0.94 to −0.04)	−0.83 (−1.49 to −0.17)	−0.76 (−1.26 to −0.25)	−0.93 (−1.36 to −0.51)	−0.53 (−1.01 to −0.06)
P value	.001	.001	**.06**	.035	<.015	<.005	<.001	.028
Number of children in house ≥3	−0.61 (−0.97 to −0.26)	−0.91 (−1.27 to −0.54)	−0.46 (−0.81 to −0.11)	−0.54 (−0.89 to −0.20)	−0.98 (−1.49 to −0.47)	−0.80 (−1.18 to −0.42)	−0.74 (−1.07 to −0.42)	−0.39 (−0.71 to −0.06)
P value	.001	<.001	.011	.003	<.001	<.001	<.001	.021
Age at implantation ≥4	−0.59 (−0.93 to −0.26)	−0.82 (−1.18 to −0.47)	−0.42 (−0.76 to −0.08)	−0.59 (−0.93 to −0.26)	−1.05 (−1.54 to −0.55)	−0.83 (−1.19 to −0.46)	−0.77 (−1.08 to −0.45)	−0.47 (−0.79 to −0.15)
P value	.001	<.001	.015	.001	<.001	<.001	<.001	.005
Duration of implant use ≥5	−0.75 (−1.20 to −0.31)	−0.74 (−1.18 to −0.29)	**−0.41 (−0.90 to 0.08)**	−0.46 (−0.87 to −0.05)	−0.78 (−1.33 to −0.23)	−0.72 (−1.21 to −0.24)	−0.90 (−1.29 to −0.50)	−0.51 (−0.98 to −0.05)
P value	.002	.002	**.10**	.031	<.007	<.005	<.001	.032

[a]For "no covariates," differences in the group means seen in Table 2 are reported, along with the Wilcoxon rank-sum test *P* values repeated from **Table 2**. For "all covariates," the analysis of variance (ANOVA) model includes all the covariates listed in the table, while the remaining rows consider ANOVA models with just the single specified covariate. The estimated differences in mean domain scores are based on the covariate adjustments. Values in bold did not reach significance.

it has been the survey that has been used in the most recent studies.[4,10,22-25]

This leads to the conclusion that although the survey is not flawless, it is reasonable to use as a tool to understand parental perspectives for children with CI. In the absence of any other HRQoL surveys for NCNIDD, this survey does attain meaningful data. In this study, parents of normally developing children responded more favorably to CI in all domains in comparison to the children with NCNIDD. When looking at children with NCNIDD, parents overall did respond neutrally or slightly favorably to all domains except ''support the child'' and ''self-reliance'' domains. This suggests that parents of children with NCNIDD do perceive benefits to CI in terms of quality of life, but they are more strongly perceived by parents of normally developing children.

There are currently limited surveys that are specifically created for children with developmental delays, even in general pediatric use. Previous studies have shown that parents report lower HRQoL scores in comparison to parents of normative peers for other cognitive disorders in general,[27] including cerebral palsy[28] and Down syndrome.[29] The surveys used in the above studies, Child Health Questionnaire and Kidscreen-52, are meant for use with healthy children and those with both acute and chronic health conditions. Our study was done with the best available survey for children with cochlear implants, but it highlighted the limitations of using these surveys in children with developmental delays and identified a gap in literature. The potential full benefits of CI may not have been captured because the parents are answering on basis of the underlying neurological impairment. This is a limitation with the use of a survey not devised for children with disabilities, and it is unclear if the parents of children with NCNIDD were able to answer the survey based on the impact of CI and not allow the underlying neurological impairment flaw the responses. This underscores, however, the need for an improved construct to evaluate condition-specific HRQoL in CI patients, and even more so specific to children with disabilities. Developing and validating a survey for use in these patients would be essential to understand the true impact.

Socioeconomic and family-related variables have previously been shown to affect HRQoL assessments in past. Previous studies have reported improved outcomes for QoL measures in females, older children, higher parental occupation skill level, age of CI at less than 5 years, and use of CI longer than 4 years.[30,31] However, other studies showed that there were no correlations or an inverse relationship between age and duration of CI experience and the domains.[4,32] Within this study, age at implantation and duration of CI experience had no impact on mean differences in domain scores except in the ''effects of implantation'' domain.

There was loss of significance in ''effects of implantation'' when adjusted for education (no change in magnitude of group difference) and duration of implant use (change of

0.05 in group difference). In addition, adjusting for socioeconomic and family variables, comparisons of normally developing children to children with NCNIDD were of similar magnitude and statistical significance to the unadjusted comparisons in nearly all domains. In ''support the child'' and ''well-being and happiness'' domains, there was a loss in statistical significance, but magnitude in the group difference upon covariate adjustment was only 0.02 and 0.03, respectively. With the minimal changes in the magnitude of group differences in all these cases of loss of statistical significance, it is unclear if the loss of significance is a true reflection of impact of the variables or due to small numbers of patients in the cohorts. Further studies with larger cohorts could help elucidate if these socioeconomic and familial variables do affect perception of HRQoL benefits in these patients.

Other studies that evaluated quality of life in developmentally delayed children have used varied definitions, all highlighting variation in physical or mental abilities. One article defined the developmentally delayed group as ''IQ in the moderate, severe or profound range, with or without autism spectrum disorder (ASD), physical disabilities and mental health/behavioral issues.''[33] Another article took all children with unclassified developmental delays, which were broken down into 6 categories: gross-motor delays, fine-motor delays, speech and language delays, cognitive delays, behavioral and psychological delays, and global delays.[34] Quantitative measures that also can assess developmental delays include IQ testing, Peabody Developmental Motor Scales, Gross Motor Function Measure, Preschool Language Evaluation Tool, Child Expression Evaluation Tool, Chinese Wechsler Intelligence Scale for Children (third edition), and Baley II Scales of Infant and Toddler Development.[34]

The inclusion criteria for the NCNIDD group were designed to select the most significantly neurologically affected children as possible, encompassing the limitations mentioned in various ways in other studies: disability in cognition, communication, and mental health/behavioral issues. Unfortunately, the results of quantitative testing of the patients prior to study enrollment were not available. However, all of the patients in the group had been diagnosed with developmental delay by a neurologist or developmental pediatrician, presumably using these quantitative metrics. These highly selective criteria limited the number of patients in the study group, but they best reflected the patient population that the study question was to address. Despite the small group sample, the study was able to detect differences between the 2 groups. One weakness of the study is the differences in cohorts in terms of number of children in household and race. The number of children in the household was higher in the NCNIDD group, and in terms of race, there was a higher percentage of white families in the normally developing group in comparison to the NCNIDD group. Age and race, however, were among the covariates for which adjustments were made using ANOVA to compare the groups with respect to mean domain scores.

As with any survey studies, recall bias is a weakness within this study, with reliance on responses from parents of children with an average age of duration of implantation at nearly 5 years. Last, low response rates often plague studies of this kind in which patients are surveyed. One study evaluated 490 studies in 17 refereed academic journals, and the mean (SD) response rate was 52.7% (20.4%) for studies that used data collected from individuals and 35.7% (18.8%) for studies that used data collected from organizations.[35] Our response rates did fall within 1 SD of these ranges and is within the range of usual return rates quoted in literature. In addition, with no differences in duration of implantation and survey response rates from both groups, it is anticipated these biases would affect both groups similarly.

Conclusions

In this study of HRQoL with CI, parents of normally developing children responded more favorably toward all domains in comparison to children with NCNIDD. Differences between groups in mean domain scores, univariably and almost universally when adjusting for socioeconomic and family variables individually and collectively, were statistically significant. Parents of children with NCNIDD did respond neutral or favorably toward most domains, albeit less strongly than normally developing children. This suggests that there are HRQoL benefits of CI perceived by parents of both the patients with NCNIDD and normally developing patients, even in a setting of limited cognitive and physical functioning. Surveys tailored to evaluate patients with cognitive and developmental disabilities would be useful in further understanding the HRQoL of patients with NCNIDD who undergo cochlear implantation.

Acknowledgments

We thank Jeff Hammel, MS, for assistance with data analysis and John O'Neill, research coordinator for the Head and Neck Institute at the Cleveland Clinic, for assistance for data collation.

Author Contributions

Samantha Anne, substantial contributions to conception and design, acquisition of data, analysis and interpretation of data, acknowledgment of drafting the article and revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved; **Seth Roslow Schwartz**, substantial contributions to conception and design, analysis and interpretation of data, acknowledgment of drafting the article and revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved; **Jennifer L. McCoy**, acquisition of data, analysis and interpretation of data, acknowledgment of drafting the article and revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to

the accuracy or integrity of any part of the work are appropriately investigated and resolved; **Thomas Haberkamp**, substantial contributions to conception and design, analysis and interpretation of data, acknowledgment of drafting the article and revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved; **Michael E. Hoffer**, analysis and interpretation of data, acknowledgment of drafting the article and revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved; **David H. Chi**, substantial contributions to conception and design, analysis and interpretation of data, acknowledgment of drafting the article and revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Disclosures

Competing interests: None.

Sponsorships: None.

Funding source: None.

Supplemental Material

Additional supporting information is available in the online version of the article.

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