

ONLINE FIRST

Sequential Bilateral Cochlear Implantation in Children

Quality of Life

Marloes Sparreboom, MA; Ad F. M. Snik, PhD; Emmanuel A. M. Mylanus, MD, PhD

Objective: To assess the effect of sequential bilateral cochlear implantation in children on their quality of life (QoL).

Design: Prospective cohort-control study.

Setting: Tertiary academic referral center.

Patients: Thirty children with prelingual deafness underwent sequential bilateral cochlear implantation (mean age at first implant, 1.8 years; mean age at second implant, 5.3 years). Nine children with a unilateral cochlear implant were also included in the study. All children had prelingual deafness, had good implant performance, and had no benefit from a contralateral conventional hearing aid. The groups were matched on duration of unilateral implant use, chronological age, and degree of bilateral hearing loss.

Intervention: Sequential bilateral cochlear implantation.

Main Outcome Measures: Quality of life was assessed before surgery of the second implant and after 12 and 24 months of bilateral implant use. In the children with a unilateral cochlear implant, QoL was assessed over time and was compared with the study group after 12 and 24 months. Six questionnaires were used to measure QoL: overall health status using a visual analog scale; the Health Utilities Index Mark 3 (HUI3); the Pediatric Quality of

Life Inventory (PedsQL); the Glasgow Children's Benefit Inventory (GCBI); the Speech, Spatial, and Qualities of Hearing Scale (SSQ); and the Nijmegen Cochlear Implant Questionnaire (NCIQ).

Results: Results showed no significant gain in generic QoL measures associated with sequential bilateral cochlear implantation ($P > .05$). The nonsignificant effect yielded on the HUI3 could be attributed to the ceiling effect and the lack of resolution in the hearing domain obtained in cochlear implant recipients. The gain in QoL of +0.04 may, therefore, be underestimated. In contrast, the 3 disease-specific questionnaires showed a significant improvement in QoL ($P < .05$). Results also showed that, unlike the children with a unilateral implant, QoL measures continued to improve with longer durations of bilateral implant use. Within the study group, the age at second implantation had no influence on the gain in QoL ($P > .05$).

Conclusion: Sequential bilateral cochlear implantation in children is associated with an improvement in QoL, although this is predominantly reflected in the disease-related aspects of QoL and not necessarily in generic QoL.

Arch Otolaryngol Head Neck Surg. 2012;138(2):134-141.
Published online January 16, 2012.
doi:10.1001/archoto.2011.229

Author Affiliations: Radboud University Nijmegen Medical Centre, Department of Otorhinolaryngology, Head and Neck Surgery, Hearing and Implants, Donders Institute for Brain Cognition and Behaviour, Nijmegen, the Netherlands.

NOWADAYS, IN A LIMITED number of countries, bilateral cochlear implantation in children is reimbursed. Because most studies in children indicate that speech perception in noise and sound localization are better with bilateral cochlear implants (BiCIs) compared with a unilateral cochlear implant (UCI),¹ children with BiCIs will presumably acquire bilateral advantages in everyday situations. Most studies investigating children with BiCIs focus on outcomes in speech perception and sound localization. Because results collected in clinical settings are not always representative of everyday situations,² it

is also of importance to gather information about the quality of life (QoL) for these children.

Quality of life can be measured by the use of generic or disease-specific questionnaires. For studies in children, parent-proxy questionnaires are most commonly used. The advantages of generic questionnaires are that they cover a broad range of QoL dimensions, and the outcomes can be compared with other interventions. Owing to this broad approach, generic questionnaires may show less responsiveness to the effects of an intervention.³ Disease-specific questionnaires, however, only include domains that apply to the disease but cannot be compared with

other interventions. Therefore, it is important to include disease-specific questionnaires as well as generic questionnaires.³ In children with BiCIs, only a few studies have reported on QoL.⁴⁻⁷

Scherf et al⁷ reported on disease-specific instruments, namely the Categories of Auditory Performance, the Speech Intelligibility Rating, and the questionnaire. Scherf et al⁷ stratified their participants into 2 age groups: children who received the second cochlear implant (CI2) before the age of 6 years and children who received the CI2 at age 6 years or older. After 36 months of BiCI use, the number of children who obtained higher Categories of Auditory Performance and Speech Intelligibility Rating scores increased significantly. The Würzburg questionnaire showed that the younger children had more positive bilateral experiences and less negative bilateral experiences compared with the older children. However, after 36 months of CI2 use, no significant differences were found in the negative bilateral experiences between the 2 groups.

In a study by Lovett et al,⁶ 3 separate questionnaires were used: the disease-specific Speech, Spatial, and Qualities of Hearing Scale for parents (SSQ); the Health Utilities Index Mark 3 (HUI3); and a generic QoL questionnaire using a visual analog scale (VAS). Parents of children with a UCI and parents of children with BiCIs completed the questionnaires. Lovett et al⁶ reported that except for the speech and spatial domains on the SSQ, there were no significant differences between the parental ratings of the UCI and the BiCI groups. For the speech and spatial domains, the parental ratings of the BiCI group were significantly higher than the UCI group. In agreement, Beijen et al⁴ and Galvin et al⁵ found that children with BiCIs scored higher on the spatial domain of the SSQ after bilateral implantation⁵ or compared with a control group of children with a UCI.⁴ Beijen et al⁴ also reported on a generic QoL questionnaire—the Pediatric Quality of Life Inventory (PedsQL). Like Lovett et al,⁶ the 2 cochlear implant groups of Beijen et al⁴ did not show significant differences in generic QoL. To our knowledge, besides Lovett et al⁶ and Beijen et al,⁴ no further studies have examined generic QoL in children with BiCIs. A limitation is that these studies were not longitudinal. Scherf et al⁷ showed that bilateral advantages increase over time. Especially in children with BiCIs that are implanted sequentially, it is conceivable that bilateral advantages require more time to develop as the newly implanted side has to integrate with the experienced cochlear implant side.

In 2006, we started a longitudinal study to assess the effects of sequential bilateral cochlear implantation in 30 children on electrophysiological outcomes, primary outcomes of bilateral hearing and outcomes on QoL. Prior reports from this study already showed significant benefits in speech perception and sound.⁸ Improvements over time were also apparent in longitudinal electrophysiological measures.⁹ The present study supplements the previous reports by focusing on QoL. Quality of life in the children with BiCIs was assessed longitudinally, and results were also compared over time with a reference group of 9 children with a UCI. To measure QoL, parents completed

generic as well as disease-specific questionnaires. The influence of age in receiving the CI2 on the size of the bilateral advantage in QoL was assessed. To assess if disease-specific QoL was in agreement with the laboratory performance, results on disease-specific QoL were correlated with the results on speech perception in noise and lateralization scores, obtained in a previous report by us.⁸

METHODS

SUBJECTS

BiCI Group

The 30 children in our study group were implanted at the Radboud University Nijmegen Medical Centre and received sequential BiCIs (Nucleus 24 multichannel devices; Cochlear Corp Australia) between April 2006 and June 2007 (referred to as the BiCI group). The eligibility criteria for bilateral implantation were as follows: at least 1 year of UCI experience; a bilateral profound prelingual hearing loss; implanted with a UCI before the age of 3 years, in which a full insertion of the electrode array was achieved; a chronological age less than 9.0 years; no benefit from a contralateral conventional hearing aid; no cognitive, learning, and/or behavioral deficits; no progressive systemic disease; no anatomical malformations that might prevent full insertion of electrode array or that might stimulate the facial nerve; and no medical condition that contraindicates the use of general anesthesia during the implant procedure. All parents were acquainted with the protocol of the study, and written informed consent was obtained before their child received the CI2. The Committee on Research Involving Human Subjects of the Radboud University Nijmegen Medical Centre approved testing.

Median age at time of CI1 implantation was 2.0 years (25th and 75th percentiles, 1.3-2.2 years [range, 0.9-2.7 years]), while the median age at CI2 implantation was 5.0 years (25th and 75th percentiles, 3.8-6.7 years [range, 2.4-8.5 years]). The median interimplant interval was 3.3 years (25th and 75th percentiles, 2.5-4.5 years [range, 1.2-7.2 years]). All the children had congenital deafness except 3 who had acquired prelingual deafness as a result of meningitis. Full insertion of the electrode array into the cochlea was achieved in all cases. All the children were able to obtain speech perception with their CI2 (full details were previously described by Sparreboom et al⁸).

UCI Group

To confirm that QoL would not change over time by the cause of maturation, a group of 9 children with a UCI (Nucleus 24 multichannel devices) was included. These 9 children (referred to as the UCI group) were matched with the BiCI group for (1) age at unilateral implantation, (2) chronological age, and (3) degree of bilateral hearing loss. All the children were prelingually deaf and did not have any diagnosed cognitive, learning, and/or behavioral deficits. They had received their cochlear implant at a median age of 1.6 years (25th and 75th percentiles, 1.3-1.9 years [range, 0.8-2.0 years]). Full insertion of the electrode array into the cochlea was achieved in all cases. It is important to realize that all the subjects in the UCI group fell within the eligibility criteria for bilateral implantation, but the parents of these children had decided not to proceed with CI2 surgery for various reasons (eg, the risk of affecting the vestibular system due to im-

plantation and the burden of the operation and rehabilitation on the child).

QUESTIONNAIRES

The parents of the study group completed the questionnaires on 3 occasions—before CI2 surgery (QoL assessment 1) and after 12 and 24 months of BiCI use (QoL assessments 2 and 3, respectively). The parents of the children with a UCI only completed the questionnaires twice with a 1-year interval between the 2 questionnaires. These 2 results for the UCI group were compared with the study group's results at QoL assessments 2 and 3. Questionnaires were distributed and explained to the parents by a single person. Parents completed all questionnaires at home and were not able to refer to their previous answers.

GENERIC QUESTIONNAIRES

Parents indicated the overall health status of their child using a VAS, which ranged from 0 (death) to 10 (perfect health). To aid questionnaire comparison, VAS scores were divided by 10.

Generic QoL was also assessed with the HUI3,¹⁰ which consists of 8 dimensions of health status. Although the parent-proxy version of the HUI3 has been validated for children of 5 years and older,¹¹ parents of children at the age of 4 years also completed the HUI3. The HUI3 scores were transformed into a utility function, in which 0 corresponds with "death" and 1 with "perfect health."

Parents also completed the parent-proxy version of the PedsQL 4.0,¹² in which health related QoL is measured on 4 domains. For the children aged 5 years and older who were able to complete the child self-report, the PedsQL for children was examined as well. The PedsQL was completed by an experienced pediatric clinician together with the child. Ratings were given on a 5-point Likert scale, ranging from "never a problem" to "almost always a problem," resulting in a score ranging from 0 to 100. For the children aged 5 to 7 years, ratings were given on a 3-point Likert scale. For comparison purposes, PedsQL scores were divided by 100.

DISEASE-SPECIFIC QUESTIONNAIRES

The Glasgow Children's Benefit Inventory (GCBI)¹³ was filled in once by parents of the BiCI group after 24 months. No GCBI scores were gathered by parents of the children with a UCI. The GCBI can be used to measure and evaluate a child's health benefit retrospectively, after an otological intervention. Answers are provided on a 5-point Likert scale ranging from "much better" to "much worse." Scores were then converted and ranged from -100 (maximum harm) to +100 (maximum benefit). The GCBI can also be analyzed per domain: emotion, physical health, learning, and vitality.¹³

Parents also completed the SSQ developed by Gatehouse and Noble.¹⁴ We used the parent-proxy version as adjusted by Galvin et al⁹ and translated into Dutch by the Katholieke Universiteit Leuven, ExpORL, Belgium. The SSQ can be subdivided into 3 sections: speech, spatial, and qualities of hearing. The child's performance is given on a VAS ranging from 0 (not at all) to 10 (perfectly). For comparison purposes, SSQ scores were divided by 10.

Furthermore, the Nijmegen Cochlear Implant Questionnaire (NCIQ) was administered. The NCIQ addresses 6 domains: basic sound perception, advanced sound perception, speech production, self-esteem, activity limitations, and social interactions.¹⁵ Answers are provided on a 5-point Likert scale, which can be transposed into scores ranging from 0 (very poor) to 100 (optimal). For comparison purposes, NCIQ scores were divided by 100.

BEHAVIORAL MEASURES

Behavioral measures were presented in Sparreboom et al.⁸ In short, speech perception in noise was measured with the Dutch Automated Toy Discrimination Test (Dutch ATT),^{16,17} in which a speech reception threshold was obtained. A 60-dB sound pressure level fixed speech-shaped noise was added at a (-)90° azimuth on the CI1.

Lateralization was measured by the use of an adaptive left-right discrimination test set-up—the minimum audible angle (MAA). The used stimuli were prerecorded and filtered (0.5-4.0 kHz) common children's songs at a fixed 65-dB sound pressure level. Full details were previously reported by us.⁸

DATA ANALYSES

Statistical analyses were performed using SPSS 17.0 for Windows. In each analysis, the level of statistical significance was set at an α level of .05. The results of the children with BiCIs after 12 and 24 months of BiCI use were compared with their preoperative measures using the Wilcoxon signed rank test for related samples. The results after 12 and 24 months of BiCI experience were compared with the results of the UCI group using independent-samples Mann-Whitney tests. Because the GCBI is a retrospective questionnaire, we tested if the group results differed significantly from the median of 0 (no effect) using a 1-sample Wilcoxon signed rank test. When statistical differences were identified (within-subjects analysis, between-subjects analysis, or both), questionnaire domains were also analyzed. To compare changes over time in the BiCI group, the Friedman analysis of variance test for related samples was used. The Wilcoxon signed rank test was used as a post hoc test and Bonferroni correction was applied to critical values. If significant bilateral advantages were found, we also investigated the correlation between age at CI1 and CI2 with the questionnaire ratings (Spearman correlation coefficient). To compare changes over time in the UCI group, we used the Wilcoxon signed rank test for related samples. For the BiCI group, Spearman correlation coefficients were calculated between disease-specific questionnaires and (1) speech perception in noise and (2) the MAA.

RESULTS

Questionnaires were returned for all children with the exception after 12 months of BiCI use, at which the parents of 1 child from the BiCI group did not return all the questionnaires. **Table 1** and **Table 2** present the descriptive statistics for each questionnaire per subject group at each measurement point.

GENERIC QUESTIONNAIRES

None of the generic questionnaires showed a significant gain in QoL after bilateral cochlear implantation ($P > .05$) (Table 1).

The PedsQL ratings provided by the parents (parent-proxy rating) were compared with the child self-reports. No significant differences in ratings were found at all measurement times for the children with BiCIs (preoperative, $z = -1.9$ [$P = .06$]; 12 months, $z = -1.5$ [$P = .13$]; and 24 months, $z = -1.6$ [$P = .12$]), as well as for the children with a UCI (12 months, $z = -0.1$ [$P = .92$]; and 24 months, $z = -1.5$ [$P = .13$]). We, therefore, only present the parent-proxy PedsQL in Table 1.

Table 1. Descriptive Statistics for Bilateral Cochlear Implant Group Between Preoperative and Postoperative Assessments^a

Questionnaire	Preoperative				After 12 mo				After 24 mo			
	Median		No.		Median		No.		Median		No.	
					Wilcoxon Signed Rank Test				Wilcoxon Signed Rank Test			
					z				P Value			
					z				P Value			
VAS	0.90 (0.80-0.95)	30	0.90 (0.80-0.97)	30	-0.3	.74	0.90 (0.80-0.95)	30	0.90 (0.80-0.96)	30	-0.4	.66
HUI3	0.58 (0.53-0.78)	23	0.66 (0.53-0.79)	23	-1.9	.06	0.58 (0.53-0.78)	22	0.76 (0.57-0.82)	22	-1.2	.24
PedsQL	0.85 (0.78-0.89)	29	0.81 (0.72-0.90)	29	-0.5	.60	0.84 (0.79-0.89)	30	0.82 (0.67-0.89)	30	-1.0	.30
GCBI	NA	NA	NA	NA	NA	NA	NA	NA	10.42 (5.73-32.29)	30	-4.2	<.001
SSQ	0.48 (0.39-0.58)	29	0.60 (0.51-0.66)	29	-3.8	<.001	0.49 (0.39-0.59)	30	0.62 (0.56-0.72)	30	-4.2	<.001
NCIQ	0.74 (0.66-0.82)	29	0.78 (0.69-0.84)	29	-1.9	.06	0.74 (0.66-0.82)	29	0.79 (0.71-0.87)	29	-2.4	.02

Abbreviations: GCBI, Glasgow Children's Benefit Inventory; HUI3, Health Utilities Index Mark 3; NA, not applicable; NCIQ, Nijmegen Cochlear Implant Questionnaire; PedsQL, Pediatric Quality of Life Inventory; SSQ, Speech, Spatial, and Qualities of Hearing Scale; VAS, visual analog scale.

^aMedians and 25th and 75th percentiles (in parentheses) were calculated within the children with BiCIs between postoperative and preoperative assessments. Wilcoxon signed rank statistics are also listed. Statistically significant differences are displayed in bold.

Table 2. Descriptive Statistics for the 2 Cochlear Implant Groups at the Second and Third QoL Assessment^a

Questionnaire	QoL 2 Assessment						QoL 3 Assessment					
	UCI Group		BiCI Group		Mann-Whitney Test		UCI Group		BiCI Group		Mann-Whitney Test	
	Median	No.	Median	No.	z	P Value	Median	No.	Median	No.	z	P Value
VAS	0.90 (0.90-1.00)	9	0.90 (0.80-0.97)	30	-1.2	.23	1.00 (0.90-1.00)	9	0.90 (0.80-0.96)	30	-1.7	.10
HUI3	0.71 (0.58-0.78)	8	0.66 (0.53-0.78)	28	-0.9	.39	0.71 (0.62-0.82)	8	0.74 (0.55-0.82)	28	-0.9	.37
PedsQL	0.84 (0.69-0.94)	9	0.81 (0.72-0.90)	29	-1.6	.10	0.88 (0.69-0.94)	9	0.82 (0.67-0.89)	30	-0.6	.57
SSQ	0.56 (0.43-0.65)	9	0.60 (0.51-0.66)	29	-0.4	.67	0.50 (0.43-0.65)	9	0.62 (0.56-0.72)	30	-2.1	.04
NCIQ	0.84 (0.71-0.88)	9	0.78 (0.69-0.84)	29	-1.0	.31	0.81 (0.71-0.89)	9	0.79 (0.71-0.87)	30	-0.9	.38

Abbreviations: BiCIs, bilateral cochlear implants; GCBI, Glasgow Children's Benefit Inventory; HUI3, Health Utilities Index Mark 3; NCIQ, Nijmegen Cochlear Implant Questionnaire; PedsQL, Pediatric Quality of Life Inventory; QoL, quality of life; SSQ, Speech, Spatial, and Qualities of Hearing Scale; UCI, unilateral cochlear implant; VAS, visual analog scale.

^aMedians and 25th and 75th percentiles (in parentheses) were calculated between children with BiCIs and a UCI at the second and third QoL assessment. Mann-Whitney statistics are also listed. Statistically significant differences are displayed in bold.

Table 3. Descriptive Statistics for the Glasgow Children's Benefit Inventory Domains^a

Domain	After 24 mo		Wilcoxon Signed Rank Test	
	Median	No.	z	P Value
Emotion	4.17 (0.00-33.33)	30	-2.8	.005
Physical health	10.72 (0.00-16.08)	30	-3.4	.001
Learning	22.92 (8.33-44.80)	30	-4.2	<.001
Vitality	10.00 (3.75-31.25)	30	-3.7	<.001

^aMedians and 25th and 75th percentiles (in parentheses) were calculated. The 1-sample Wilcoxon signed rank statistics are also listed. Statistically significant differences are displayed in bold.

DISEASE-SPECIFIC QUESTIONNAIRES

The GCBI showed a significant gain in QoL after bilateral implantation ($z = -4.2$ [$P < .001$]). Within-subjects analyses of SSQ scores showed significantly higher ratings after 12 and 24 months of BiCI use compared with preoperative ratings (12 months, $z = -3.8$ [$P < .001$]; and 24 months, $z = -4.2$ [$P < .001$]). Between-subjects analyses showed a similar pattern after 24 months of BiCI use (12 months, $U = 118.0$ [$P = .67$]; and 24 months, $U = 73.0$ [$P = .04$]). The NCIQ showed significant improvements

in the within-subjects analysis (12 months, $z = -1.9$ [$P = .06$]; and 24 months, $z = -2.4$ [$P = .02$]), but no significant differences were observed in the between-subjects analysis (12 months, $U = 101.0$ [$P = .31$]; and 24 months, $U = 105.0$ [$P = .38$]).

The GCBI

All 4 GCBI domains significantly differed from zero (**Table 3**). Ratings on the 4 domains differed significantly ($\chi^2 = 41.6$ [$P < .001$]). Post hoc tests showed that

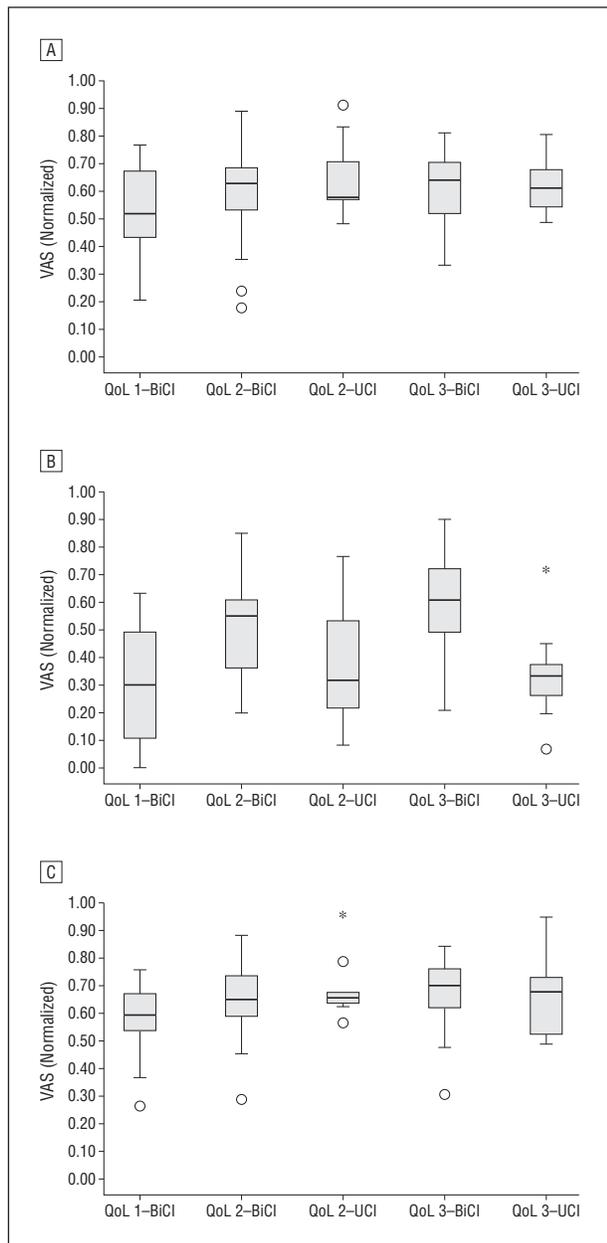


Figure. Box plots with regard to the parental ratings of the domains of the Speech, Spatial, and Qualities of Hearing Scale for speech (A), spatial (B), and qualities of hearing (C). Boxes present the median and the 25th and 75th percentiles. The end of the whiskers present the maximum and minimum values. Outliers are indicated with circles (1.5-3.0 times the interquartile range) or asterisks (>3 times the interquartile range). Box plots are plotted for the bilateral cochlear implant (BiCI) and unilateral cochlear implant (UCI) groups over time. QoL indicates quality of life; VAS, visual analog scale.

the learning domain was rated significantly higher than the other domains ($P < .008$).

The SSQ

The **Figure** presents the results for the 3 separate domains of the SSQ. When we compared the within-subjects SSQ ratings, all domains showed significant bilateral advantages after 24 months (speech, $z = -2.1$ [$P < .05$]; spatial, $z = -4.6$ [$P < .001$]; and qualities, $z = -3.3$ [$P < .01$]). The SSQ between subjects only showed sig-

nificantly higher ratings on the spatial domain for the BiCI group as compared to the UCI group after 24 months (speech, $U = 131.0$ [$P = .89$]; spatial, $U = 37.0$ [$P < .01$]; and qualities, $U = 109.5$ [$P = .40$]).

Duration of BiCI use had a significant positive effect on ratings ($\chi^2 = 28.9$ [$P < .001$]). Post hoc comparisons showed that significant improvements were seen at all measurement points after bilateral implantation ($P < .02$). For the UCI group, SSQ ratings did not improve over time ($z = -0.8$ [$P = .44$]).

The NCIQ

Table 4 gives the results for the NCIQ domains. As no differences were observed between subjects ($P > .05$) we only investigated the domains from the BiCI group. After 12 months, significant bilateral advantages were only found in the domain “advanced sound perception” ($z = -2.1$ [$P = .04$]). After 24 months, significant bilateral advantages were also present in the domains “speech production” ($z = -3.1$ [$P = .002$]) and “activity limitations” ($z = -2.4$ [$P = .01$]).

In the BiCI group, NCIQ ratings improved significantly over time ($\chi^2 = 10.8$ [$P < .01$]). Compared with preoperative measures, post hoc test results revealed that significant improvements in QoL were only seen after 24 months of BiCI use ($P < .002$). In the UCI group, no significant improvement was seen over time ($z = -0.8$ [$P = .44$]).

FACTORS OF INFLUENCE AND BEHAVIORAL MEASURES

Correlation coefficients were calculated between questionnaire ratings indicating a significant bilateral advantage (GCBI, SSQ, and NCIQ) and age at CI1 and CI2. No correlations were found between age at either implantation and ratings on the GCBI, the SSQ and the NCIQ ($P > .05$).

At QoL assessment 3, for speech perception in noise and the MAA, a clear bilateral advantage was found in comparison with the CI1 alone condition.⁸ Correlation coefficients between speech perception in noise with BiCIs and disease-specific questionnaire ratings after 24 months (GCBI, SSQ, or NCIQ) showed no significant correlations (GCBI, $r[29] = 0.132$ [$P = .50$]; SSQ, $r[29] = 0.030$ [$P = .88$]; and NCIQ, $r[28] = 0.020$ [$P = .92$]). Correlation coefficients between the MAA and either the GCBI or the NCIQ also did not show any significant correlations (GCBI, $r[24] = -0.391$ [$P < .06$]; and NCIQ, $r[23] = -0.268$ [$P = .22$]). A significant inverse correlation was found between the MAA and rating on the SSQ ($r[24] = -0.416$ [$P < .05$]), suggesting that better MAA thresholds lead to improved ratings on the SSQ.

COMMENT

This study presented data on the effect of sequential bilateral cochlear implantation on the QoL of 30 children with profound sensorineural hearing loss. Quality of life was assessed prior to CI2 surgery and after 12 and 24 months of BiCI use. Quality of life was also assessed over

Table 4. Descriptive Statistics for the Nijmegen Cochlear Implant Questionnaire Domains Over Time^a

Statistic	Preoperative		After 12 mo		Wilcoxon Signed Rank Test		Preoperative		After 24 mo		Wilcoxon Signed Rank Test	
	Median	No.	Median	No.	z	P Value	Median	No.	Median	No.	z	P Value
	Basic sound perception	0.70 (0.61-0.79)	29	0.73 (0.63-0.86)	29	-1.6	.11	0.70 (0.61-0.79)	29	0.78 (0.63-0.86)	29	-1.7
Advanced sound perception	0.57 (0.46-0.71)	28	0.65 (0.56-0.78)	28	-2.1	.04	0.57 (0.46-0.71)	28	0.65 (0.53-0.78)	28	-2.2	.03
Speech production	0.83 (0.63-0.89)	29	0.85 (0.70-0.93)	29	-1.8	.07	0.83 (0.63-0.89)	29	0.90 (0.76-0.95)	29	-3.1	.002
Self-esteem	0.81 (0.72-0.88)	27	0.78 (0.73-0.88)	27	-0.2	.82	0.79 (0.72-0.88)	28	0.80 (0.73-0.88)	28	-0.1	.94
Activity limitations	0.82 (0.63-0.89)	27	0.81 (0.72-0.91)	27	-1.2	.25	0.82 (0.63-0.89)	27	0.84 (0.81-0.94)	27	-2.4	.01
Social interactions	0.81 (0.70-0.88)	29	0.80 (0.72-0.90)	29	-0.6	.53	0.81 (0.70-0.88)	29	0.83 (0.76-0.90)	29	-1.3	.21

^aMedians and 25th and 75th percentiles (in parentheses) were calculated within the children with bilateral cochlear implants between postoperative and preoperative assessments. Wilcoxon signed rank statistics are also listed. Statistically significant differences are displayed in bold.

time in a group of 9 children with a UCI. Results indicated that bilateral hearing with CIs is associated with higher disease-specific QoL than prior to CI2 surgery or compared with unilateral hearing. With regard to the generic questionnaires, no significant gain in QoL associated with bilateral cochlear implantation could be demonstrated. Disease-specific gains in QoL appeared to improve significantly over time, indicating a positive effect of duration of BiCI use.

GENERIC QUESTIONNAIRES

In accordance with other studies on children with BiCIs,^{4,6} no significant bilateral advantage in QoL was seen on the VAS, the HUI3, and the PedsQL. In agreement, Sach and Barton¹⁸ showed that deafness was not acknowledged by parents to be a health-related QoL issue. In addition, studies have shown that generic questionnaires are unable to distinguish between children with normal hearing and children with a cochlear implant.¹⁹ With regard to the PedsQL, ratings for both the BiCI and the UCI group fell within normal limits for healthy children as identified by Varni et al.¹² The same applies to the results of Beijen et al.⁴

The studies by Sach and Barton¹⁸ and Loy et al,¹⁹ however, did not report on the HUI3. Although the HUI3 seems to be a promising instrument for measuring QoL in hearing-impaired subjects, there are certain disadvantages with regard to cochlear implant recipients. A gain in utility associated with (bilateral) cochlear implantation would presumably be caused by an improvement in hearing, which should be reflected in the hearing dimension of the HUI3. The dimension of hearing contains 6 items to indicate the hearing status of the subject. However, only 3 of the 6 items are applicable for cochlear implant recipients because they are unable to hear without the use of their hearing device. Since the children within our study group could already hear with their UCI prior to CI2 surgery, only 2 items remained applicable. Many parents already rate their children with a UCI on a level 3 of the hearing dimension.²⁰ Consequently, not much gain in QoL is to be expected, even with larger samples. Because of this ceiling effect and the lack of resolution in the hearing domain, the gain in QoL may therefore

been underestimated. In agreement, Lovett et al⁶ reported on a nonsignificant difference in HUI3 scores between their BiCI and UCI groups, which showed a slight advantage in median utility scores for the BiCI group compared with the UCI group. When we compare our results with the gain in generic QoL associated with unilateral implantation as measured using the HUI3, the mean gain in QoL of +0.04 after 24 months of BiCI use was considerably smaller than the +0.17 gain after unilateral implantation.²¹ This gain of +0.17 in QoL after unilateral implantation was found in a group of children with a similar degree of deafness and age at unilateral implantation as our study group.

DISEASE-SPECIFIC QUESTIONNAIRES

In agreement with our data on speech perception and directional hearing,⁸ bilateral advantages were clearly apparent on the disease-specific instruments. The GCBI showed a significant benefit of sequential bilateral implantation. When the different domains of the GCBI were considered individually, the largest benefit was found in the learning domain. The advantage of the GCBI is that items are directly related to the intervention; this is not the case with the generic questionnaires. To our knowledge, only 1 study reported on the GCBI for children with cochlear implants. These results are published within a Dutch journal and showed that unilateral cochlear implantation is associated with a mean GCBI score of +36.2.²² This is fairly high compared with the mean score we found to be associated with sequential bilateral cochlear implantation (+16.6).

After 24 months of BiCI use, the SSQ showed significant advantages in QoL compared with the group of children with a UCI. The overall bilateral advantage was predominantly seen in the domain of spatial hearing. This is in agreement with other studies,^{4,7} although Lovett et al⁶ also found a significant bilateral benefit in the speech domain. As SSQ ratings significantly improved over time, perhaps, when subjects have obtained more BiCI experience, bilateral advantages in the domain of speech and qualities of hearing will also be found compared with children with a UCI. For speech perception in noise obtained in a clinical setting, the current BiCI group al-

ready showed a significant benefit compared with the UCI group after 24 months of BiCI use.⁸

The within-subjects analysis indicated that bilateral advantages as measured using the SSQ were already present after 12 months of BiCI use. These advantages were not restricted to the spatial domain but were also present in the speech domain (after 24 months) and in the qualities of hearing domain (after 12 and 24 months). These everyday situations, as measured by the SSQ, were in agreement with our results on speech perception in quiet and noise and sound source lateralization.⁸ Moreover, the results on the SSQ even correlated significantly with the MAA thresholds in the study group after 24 months of BiCI use, indicating that better MAA thresholds lead to improved disease-specific QoL in everyday life.

Unlike the SSQ, no bilateral advantages on the NCIQ were found between subjects. The NCIQ is designed for cochlear implant recipients, and therefore it aims at measuring the advantages and disadvantages associated with cochlear implantation and not necessarily with bilateral hearing. The SSQ predominantly focuses on aspects of hearing that can only be fully achieved with binaural hearing and is, therefore, sensitive to changes induced by the provision of bilateral hearing devices. For that reason, presumably, no correlations were found between the NCIQ and the behavioral measures.

FACTORS OF INFLUENCE

For children with a UCI, age at implantation has a great influence on the outcomes (eg, spoken language).²³ For children with BiCIs, this also holds true within some studies with regard to the interimplant interval or the age in receiving the CI2.¹ Within our study group, in which the CI2 was provided at the maximum age of 8.5 years, no significant effect of age in receiving the CI2 was found. This limits us to predict if this also applies to children who receive their CI2 at an older age. Another limitation of the present study was the use of parent-proxy questionnaires, especially with regard to cognitive and emotional attributes. This being said, no significant differences were found in QoL ratings between parents and their child as measured using the generic PedsQL questionnaire. However, parents may have been biased because they opted for bilateral cochlear implantation. As parents completed the questionnaires at home and were not able to refer to their previous answers, this risk of bias may have been reduced. In addition, in contrast to psychological instruments, the instruments used in the present study carry less emotional and cognitive weight.

CONCLUSIONS

Gains in QoL following bilateral cochlear implantation will predominantly be seen in more disease-specific questionnaires that contain items regarding bilateral hearing. Gains in QoL continued to improve with longer duration of BiCI use. Within our study group, after CI2 surgery, improvement in QoL was not influenced by the age at CI2. The maximum age of CI2 surgery was 8.5 years.

Because generic questionnaires are insensitive to changes in hearing status, gains in QoL as measured by these questionnaires after sequential bilateral cochlear implantation in children may be underestimated.

Submitted for Publication: February 21, 2011; final revision received July 21, 2011; accepted October 26, 2011. **Published Online:** January 16, 2012. doi:10.1001/archoto.2011.229

Correspondence: Marloes Sparreboom, MA, Radboud University Nijmegen Medical Centre, Department of Otorhinolaryngology, Head and Neck Surgery, Hearing and Implants, Donders Institute for Brain, Cognition and Behaviour, PO Box 9101, 6500 HB Nijmegen, the Netherlands (m.sparreboom@kno.umcn.nl).

Author Contributions: Ms Sparreboom had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analyses. *Study concept and design:* Sparreboom, Snik, and Mylanus. *Acquisition of data:* Sparreboom. *Analysis and interpretation of data:* Sparreboom, Snik, and Mylanus. *Drafting of the manuscript:* Sparreboom, Snik, and Mylanus. *Critical revision of the manuscript for important intellectual content:* Sparreboom, Snik, and Mylanus. *Statistical analysis:* Sparreboom. *Obtained funding:* Snik and Mylanus. *Administrative, technical, and material support:* Sparreboom and Mylanus. *Study supervision:* Snik and Mylanus. **Financial Disclosure:** None reported.

Funding/Support: This project received partial financial support from Cochlear Benelux.

Previous Presentations: This study was presented at the 11th International Conference on Cochlear Implants and Other Auditory Implantable Technologies; July 3, 2010; Stockholm, Sweden; and the 10th European Symposium on Paediatric Cochlear Implantation; May 13, 2011; Athens, Greece.

Additional Contributions: We thank all the children and their parents for their loyal and enthusiastic participation in this research project.

REFERENCES

1. Sparreboom M, van Schoonhoven J, van Zanten BG, et al. The effectiveness of bilateral cochlear implants for severe-to-profound deafness in children: a systematic review. *Otol Neurotol.* 2010;31(7):1062-1071.
2. Vidas S, Hassan R, Parnes LS. Real-life performance considerations of four pediatric multi-channel cochlear implant recipients. *J Otolaryngol.* 1992;21(6):387-393.
3. Fletcher A, Gore S, Jones D, Fitzpatrick R, Spiegelhalter D, Cox D. Quality of life measures in health care, II: design, analysis, and interpretation. *BMJ.* 1992;305(6862):1145-1148.
4. Beijin JW, Snik AF, Mylanus EA. Sound localization ability of young children with bilateral cochlear implants. *Otol Neurotol.* 2007;28(4):479-485.
5. Galvin KL, Mok M, Dowell RC. Perceptual benefit and functional outcomes for children using sequential bilateral cochlear implants. *Ear Hear.* 2007;28(4):470-482.
6. Lovett RE, Kitterick PT, Hewitt CE, Summerfield AQ. Bilateral or unilateral cochlear implantation for deaf children: an observational study. *Arch Dis Child.* 2010;95(2):107-112.
7. Scherf FW, van Deun L, van Wieringen A, et al. Functional outcome of sequential bilateral cochlear implantation in young children: 36 months postoperative results. *Int J Pediatr Otorhinolaryngol.* 2009;73(5):723-730.
8. Sparreboom M, Snik AFM, Mylanus EAM. Sequential bilateral cochlear implantation in children: development of the primary auditory abilities of bilateral stimulation. *Audiol Neurootol.* 2011;16(4):203-213.
9. Sparreboom M, Beynon AJ, Snik AF, Mylanus EA. Electrically evoked auditory

- brainstem responses in children with sequential bilateral cochlear implants. *Otol Neurotol*. 2010;31(7):1055-1061.
10. Feeny D, Furlong W, Torrance GW, et al. Multiattribute and single-attribute utility functions for the health utilities index mark 3 system. *Med Care*. 2002;40(2):113-128.
 11. Horsman J, Furlong W, Feeny D, Torrance G. The Health Utilities Index (HUI): concepts, measurement properties and applications. *Health Qual Life Outcomes*. 2003;1(1):54.
 12. Varni JW, Burwinkle TM, Seid M. The PedsQL 4.0 as a school population health measure: feasibility, reliability, and validity. *Qual Life Res*. 2006;15(2):203-215.
 13. Kubba H, Swan IR, Gatehouse S. The Glasgow Children's Benefit Inventory: a new instrument for assessing health-related benefit after an intervention. *Ann Otol Rhinol Laryngol*. 2004;113(12):980-986.
 14. Gatehouse S, Noble W. The Speech, Spatial and Qualities of Hearing Scale (SSQ). *Int J Audiol*. 2004;43(2):85-99.
 15. Hinderink JB, Krabbe PF, Van Den Broek P. Development and application of a health-related quality-of-life instrument for adults with cochlear implants: the Nijmegen cochlear implant questionnaire. *Otolaryngol Head Neck Surg*. 2000;123(6):756-765.
 16. Crul ThAM, Braks JTM, Snik AFM, Broekx JPL. Bepaling van de woordidentificatiedrempel bij kleuters. *Stem- Spraak en Taalpathologie*. 1994;3(3):161-178.
 17. Summerfield Q, Palmer AR, Foster JR, Marshall DH, Twomey T. Clinical evaluation and test-retest reliability of the IHR-McCormick Automated Toy Discrimination Test. *Br J Audiol*. 1994;28(3):165-179.
 18. Sach TH, Barton GR. Interpreting parental proxy reports of (health-related) quality of life for children with unilateral cochlear implants. *Int J Pediatr Otorhinolaryngol*. 2007;71(3):435-445.
 19. Loy B, Warner-Czyz AD, Tong L, Tobey EA, Roland PS. The children speak: an examination of the quality of life of pediatric cochlear implant users. *Otolaryngol Head Neck Surg*. 2010;142(2):247-253.
 20. Smith-Olinde L, Grosse SD, Olinde F, Martin PF, Tilford JM. Health state preference scores for children with permanent childhood hearing loss: a comparative analysis of the QWB and HUI3. *Qual Life Res*. 2008;17(6):943-953.
 21. Barton GR, Stacey PC, Fortnum HM, Summerfield AQ. Hearing-impaired children in the United Kingdom, IV: cost-effectiveness of pediatric cochlear implantation. *Ear Hear*. 2006;27(5):575-588.
 22. Hoffer MMR, Damen WJA, Hoekstra CC, Mylanus EAM. Cochleaire implantatie bij meervoudig gehandicapte kinderen: kwaliteit van leven en taalbegrip. *Stem-Spraak-en Taalpathologie*. 2006;14(2):143-160.
 23. Nicholas JG, Geers AE. Effects of early auditory experience on the spoken language of deaf children at 3 years of age. *Ear Hear*. 2006;27(3):286-298.