ARTICLE

Effect of Pediatric Bilateral Cochlear Implantation on Language Development

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Objective: To examine spoken language outcomes in children undergoing bilateral cochlear implantation compared with matched peers undergoing unilateral implantation.

Design: Case-control, frequency-matched, retrospective cross-sectional multicenter study.

Setting: Two Belgian and 3 Dutch cochlear implantation centers.

Participants: Twenty-five children with 1 cochlear implant matched with 25 children with 2 cochlear implants selected from a retrospective sample of 288 children who underwent cochlear implantation before 5 years of age.

Intervention: Cochlear implantation.

Main Outcome Measures: Performance on measures of spoken language comprehension and expression (Reynell Developmental Language Scales and Schlichting Expressive Language Test).

Results: On the receptive language tests (mean difference [95% CI], 9.4 [0.3-18.6]) and expressive language tests (15.7 [5.9-25.4] and 9.7 [1.5-17.9]), children undergoing bilateral implantation performed significantly better than those undergoing unilateral implantation. Because the 2 groups were matched with great care on 10 auditory, child, and environmental factors, the difference in performance can be mainly attributed to the bilateral implantation. A shorter interval between both implantations was related to higher standard scores. Children undergoing 2 simultaneous cochlear implantations performed better on the expressive Word Development Test than did children undergoing 2 sequential cochlear implantations.

Conclusions: The use of bilateral cochlear implants is associated with better spoken language learning. The interval between the first and second implantation correlates negatively with language scores. On expressive language development, we find an advantage for simultaneous compared with sequential implantation.

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CURRENTLY, MORE THAN half the profoundly deaf children in the United States are treated with cochlear implants.[1] Cochlear implants consist of an externally worn microphone and a microprocessor that extracts intensity, frequency, and timing cues from acoustic signals. The system transforms these acoustic cues into an electrical code. Internally, a surgically placed receiver transmits the code to an implanted electrode array that stimulates surviving auditory neurons.

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Several studies have shown that a second cochlear implant in children has a positive effect on auditory development. Children undergoing bilateral implantation demonstrate improved lateralization[2,3] and localization[2,4,5] skills using both implants compared with using only the first or the second implant. Besides a benefit in localization skills, it has been shown that bilateral implantation induces enhanced speech recognition. Most children achieve better speech recognition scores in quiet[6,7] and in noise[3,8,9] using both cochlear implants instead of one. Moreover, the advantages are greater in children with a limited interimplantation interval.[10]

Improved localization and speech recognition skills enhance the ability to perceive speech in more challenging listening environments, such as noisy classrooms and family gatherings. This improved speech perception could facilitate the ability to pick up language in everyday life. At this time, evidence on the longterm effect of bilateral cochlear implantation on language development is lacking.[11-13] This is partly because individual cochlear implantation centers have too few children undergoing the procedure to control for other variables that may influence language outcomes.

In the present multicenter study, which included 288 children with unilateral or bilateral cochlear implants, it was possible to control for several factors. We hypothesized that an augmented level of language immersion based on bilateral cochlear implantation can improve the development of language skills in these children. Therefore, receptive and expressive language outcomes were examined in children undergoing bilateral implantation compared with carefully matched peers undergoing unilateral implantation. In addition, within the group of children undergoing bilateral implantation, we evaluated the effect of the interval between the first and the second cochlear implantation on language outcome.

METHODS

PARTICIPANTS

Because 5 cochlear implantation centers (2 in Belgium and 3 in the Netherlands) participated in this multicenter study, it was possible to incorporate 288 Dutch-speaking children with unilateral or bilateral cochlear implants. The study was approved by the centers' institutional review boards and was in accordance with the tenets of the 1975 Declaration of Helsinki. Three years after implantation of the first device, language scores of 125 children were available. Twenty-five of these children from 4 cochlear implantation centers underwent bilateral implantation. None of the parents had to finance the second implantation. Sixty-four children underwent unilateral implantation and did not use a hearing aid contralaterally. The bilateral group was carefully frequency matched with children undergoing unilateral implantation. All children with bilateral implants were prelingually deaf and received their first implant before age 2.06 years. Thirty-five children met this criterion in the unilateral group. Because the bilateral group did not contain children with multiple additional disabilities, 2 children with a unilateral implant and multiple additional disabilities were excluded. All children with bilateral implants were raised in monolingual families with normal-hearing parents. Applying these criteria to the unilateral group resulted in the selection of 25 participants. The matching process is illustrated in **Figure 1**.

Every child received an implant using recent technology from Cochlear Ltd (unilateral, 20 children [40%]; bilateral, 18 [36%]) or Advanced Bionics, LLC (unilateral, 5 [10%]; bilateral, 7 [14%]). Children with bilateral implants used the same type of device in both ears. In 49 children (98%), full insertion was accomplished; 1 child (2%) had 15 usable active electrodes. Children with indications of intellectual disabilities were excluded from the study. An overview of all participants is given in **Table 1** and **Table 2**.

None of the children with a unilateral implant used a hearing aid at the nonimplanted ear. In the bilateral group, 8 children (32%) received both implants simultaneously before 2.06 years of age, whereas the others underwent sequential implantation. The second group obtained their second implant between the ages of 1.08 and 5.01 years. One child received the second implant within 1 year of the first implant. There was an equal distribution of children receiving their second implant within the second (9 children [36%]) and third (7 [28%]) year after the first one.

MATCHING CRITERIA

After selecting a unilateral implantation comparison group, both groups of children were matched on a number of factors that might influence their language development. These factors can be divided into auditory, child, and environmental factors. An overview of all variables is given in **Table 3**.

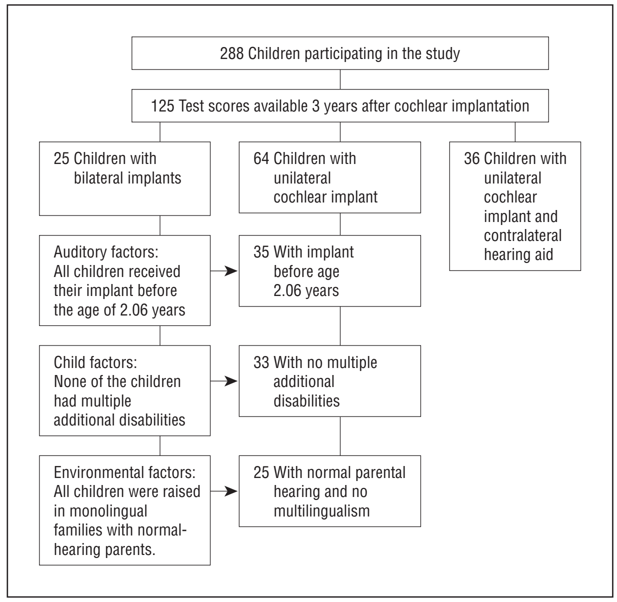

Figure 1. Flowchart illustrates the matching process between cochlear implantation groups.

The unilateral and bilateral implantation groups matched on 3 auditory properties: (1) age at first fitting, (2) age at diagnosis, and (3) the use of a hearing aid before the cochlear implantation. The 2 groups did not differ in the mean age at which the first implant was fitted (mean difference [95% CI], 2.5 [−0.3 to 5.4] mo). There was no difference in age at diagnosis between the 2 groups (mean difference [95% CI], 0.1 [−3.4 to 3.6] mo). The preimplantation use of hearing aids was comparable for the unilateral and the bilateral groups (relative risk [95% CI], 1.8 [0.7-4.9]).

The 2 groups also matched on sex, cause of deafness, and additional disabilities. Boys outnumbered girls in both groups, but the distribution was not significantly different (relative risk [95% CI], 0.7 [0.4-1.1]) between the unilateral and bilateral implantation groups. In addition, the causes of deafness were comparable for the children in both groups (likelihood ratio, 0.4; *df*, 4; $P=.98$). Finally, the presence of additional disabilities was equal in both groups (likelihood ratio, 0.2; *df*, 2; $P=.89$), including learning disorders (eg, dyslexia and dyscalculia) and motor or balance disorders.

To minimize the effect of educational environment on the language development of both groups, they were matched on 4 factors. First, the number of children attending a special school was similar between groups (relative risk [95% CI], 1.2 [0.7-2.1]). Second, all children had to be raised by hearing parents, and third, the parents were native Dutch speakers. Finally, none of the children grew up in a family with parents who were insufficiently involved in the rehabilitation process. Parental involvement was classified on the basis of observations mentioned in the child's file. Parental involvement was labeled insufficient if parents were not motivated and/or unable to fulfill their commitments (eg, not showing up for appointments).

MAIN OUTCOME MEASURES

Language outcomes on 2 standardized language tests were taken into account. Both tests were administered orally 3 years after the first implantation, between March 20, 2003, and December 8, 2009. The use of sign language by the test leader or by the child was not allowed. Children in the bilateral implanta-

Table 1. Characteristics of 25 Participants Undergoing Bilateral Cochlear Implantation

Patient No.	Timing[a]	Age at Implantation, mo		Interval, mo[b]	Age at Dx, mo	HA Use[c]	Sex	Cause of Deafness	Additional Disabilities	School
		First	Second							
1	Seq	7	20	13	1	Yes	M	Unknown	No	Regular
2	Sim	10	10	0	5	No	M	Meningitis	No	Special
3	Seq	12	26	14	10	Yes	M	Meningitis	No	Special
4	Seq	13	30	17	1	Yes	M	Unknown	Learning disability	Special
5	Seq	14	29	15	1	Yes	F	Unknown	Motor/balance disorder	Regular
6	Sim	13	13	0	10	No	M	Meningitis	No	Special
7	Sim	13	13	0	11	No	M	Meningitis	No	Regular
8	Sim	14	14	0	2	Yes	M	Meningitis	No	Regular
9	Sim	15	15	0	2	Yes	M	Prematurity	No	Regular
10	Seq	15	41	26	1	Yes	M	Unknown	No	Special
11	Sim	17	17	0	15	No	M	Meningitis	Motor/balance disorder	Special
12	Seq	16	34	18	2	Yes	M	Genetic	No	Regular
13	Sim	17	17	0	15	No	M	Meningitis	No	Regular
14	Sim	17	17	0	15	No	M	Meningitis	No	Regular
15	Seq	18	35	17	1	Yes	M	Genetic	No	Special
16	Seq	19	25	6	15	No	F	Meningitis	No	Regular
17	Seq	21	57	36	8	Yes	M	Unknown	No	Special
18	Seq	20	55	35	12	Yes	F	Genetic	No	Special
19	Seq	20	48	28	0	Yes	M	CMV	No	Special
20	Seq	22	40	18	11	Yes	M	Unknown	No	Special
21	Seq	23	45	22	12	Yes	F	Genetic	No	Regular
22	Seq	24	54	30	11	Yes	F	Genetic	No	Special
23		27	59	32	1	Yes	M	Prematurity	No	Special
24		27	59	32	15	Yes	F	Unknown	Learning disability	Regular
25		28	45	17	19	Yes	M	Genetic	No	Special

Abbreviations: CMV, cytomegalovirus; Dx, diagnosis; HA, hearing aid; Seq, sequential; Sim, simultaneous.
[a] For recipients of Seq implantation, the first implant was received before the age of 2.00 years.
[b] Indicates time from the first implantation to the second.
[c] Indicates before implantation.

tion group had at least 3 months of experience with their second implant.

The Dutch version of the Reynell Developmental Language Scales (RDLS) was used to measure the receptive language outcome.[14,15] This standardized language assessment tool is designed to be used with children aged 1.02 to 6.03 years. The test evaluates language comprehension abilities at gradually increasing levels of difficulty. At first, the vocabulary is assessed by asking the child to identify objects or pictures (eg, "Where is the ball?"). Later, the items contain small sentences with tasks that the child has to perform (eg, "Put the spoon in the cup").

The expressive counterpart of the Dutch RDLS is the Schlichting Expressive Language Test (SELT).[15,16] This test is standardized for children aged 1.09 to 6.03 years. We administered the Word Development and Sentence Development SELT subtests. In the Word Development Subtest, the expressive vocabulary skills of the children were measured by asking them to name objects or pictures. The Sentence Development Subtest evaluates the knowledge of certain syntactic structures by asking the child to repeat given sentences.

The results on both language tests were expressed as standard scores with a mean (SD) of 100 (15).

STATISTICAL ANALYSIS

Language test results were assessed by parametric analysis because the results of the bilateral and unilateral groups on the RDLS and the SELT subtests were distributed normally (Kolmogorov-Smirnov, using SPSS software, version 16.0 [SPSS Inc]). Statistical comparisons of mean values were performed using 2-tailed *t* tests for independent samples with Bonferroni correction for multiple comparisons. Statistical coherence between values was evaluated using simple linear regression analysis (Pearson correlation). Within the bilateral group, the results of the groups undergoing sequential and simultaneous implantation were not distributed normally (Kolmogorov-Smirnov). Therefore, the Mann-Whitney test for independent samples was used to assess whether a significant difference existed in standard scores of children undergoing simultaneous implantation compared with those undergoing sequential implantation. The significance criteria were established at $P < .05$.

RESULTS

Figure 2 illustrates the standard scores of the unilateral and bilateral implantation groups, an age-appropriate language level (norm), a language delay of −1 SD compared with the norm, and a delay of −2 SDs compared with the norm.

SPOKEN LANGUAGE: COMPREHENSION

With regard to the RDLS standard scores, the bilateral group (mean score, 85.6) scored significantly better than the unilateral group (mean score, 76.2) (mean difference [95% CI], 9.4 [0.3-18.6]). In the unilateral group, 12 children (48%) had a language delay of more than 2 SDs compared with the norm, whereas 2 children (8%) in the bilateral group had a delay of more than 2 SDs.

Table 2. Characteristics of 25 Participants Undergoing Unilateral Cochlear Implantation

Patient No.	Age at First Fitting, mo	Age at Dx, mo	HA Use[a]	Sex	Cause of Deafness	Additional Disabilities	School
1	9	6	Yes	M	Meningitis	Learning disability	Special
2	13	8	Yes	F	Meningitis	No	Regular
3	14	1	Yes	M	Genetic	Motor/balance disorder	Special
4	15	7	Yes	F	Unknown	Learning disability	Special
5	16	2	Yes	M	CMV	No	Special
6	17	2	Yes	M	CMV	No	Regular
7	18	15	No	M	Meningitis	No	Regular
8	18	10	Yes	M	Genetic	No	Special
9	18	1	Yes	F	Unknown	No	Regular
10	18	2	Yes	M	Genetic	No	Special
11	19	1	Yes	M	Prematurity	No	Special
12	19	15	No	F	Meningitis	No	Regular
13	21	13	No	F	Meningitis	No	Special
14	22	11	Yes	M	Meningitis	No	Special
15	22	3	Yes	F	Genetic	Learning disability	Special
16	22	10	Yes	M	Meningitis	No	Special
17	23	16	Yes	F	Meningitis	No	Regular
18	23	1	Yes	F	Unknown	No	Regular
19	24	20	Yes	M	Meningitis	No	Special
20	25	2	Yes	F	Genetic	No	Regular
21	25	15	Yes	F	Unknown	No	Special
22	26	12	Yes	F	Unknown	No	Special
23	26	15	Yes	M	Unknown	No	Regular
24	26	1	Yes	M	Prematurity	Motor/balance disorder	Special
25	26	10	Yes	M	Unknown	No	Special

Abbreviations: CMV, cytomegalovirus; Dx, diagnosis; HA, hearing aid.
[a] Indicates before implantation.

Table 3. Overview of Factors on Which the 2 Groups Matched[a]

Factor	Bilateral Group	Unilateral Group	Statistical Analysis (95% CI)
Auditory			
Age at first fitting, mean (SD), y	1.06 (0.05)	1.08 (0.05)	Mean difference, 2.5 (−0.3 to 5.4) mo
Age at diagnosis, mean (SD), y	0.08 (0.06)	0.08 (0.06)	Mean difference, 0.1 (−3.4 to 3.6) mo
HA use[b]			
Yes	18 (72)	22 (88)	Relative risk, 1.8 (0.7 to 4.9)
No	7 (28)	3 (12)	
Child			
Sex			
Male	19 (76)	14 (56)	Relative risk, 0.7 (0.4 to 1.1)
Female	6 (24)	11 (44)	
Cause of deafness			
Genetic	6 (24)	5 (20)	Likelihood ratio, 0.4; *P* = .98
CMV	1 (4)	2 (8)	
Meningitis	9 (36)	9 (36)	
Prematurity	2 (8)	2 (8)	
Unknown	7 (28)	7 (28)	
Additional disabilities			
Learning disorder	2 (8)	3 (12)	Likelihood ratio, 0.2; *P* = .89
Motor or balance disorder	2 (8)	2 (8)	
Environmental			
School			
Special	14 (56)	16 (64)	Relative risk, 1.2 (0.7 to 2.1)
Regular	11 (44)	9 (36)	
Parental hearing	Normal	Normal	
Multilingualism	No	No	
Parental involvement	Sufficient	Sufficient	

Abbreviations: CMV, cytomegalovirus; HA, hearing aid.
[a] Unless otherwise indicated, data are expressed as number (percentage) of patients.
[b] Indicates before implantation.

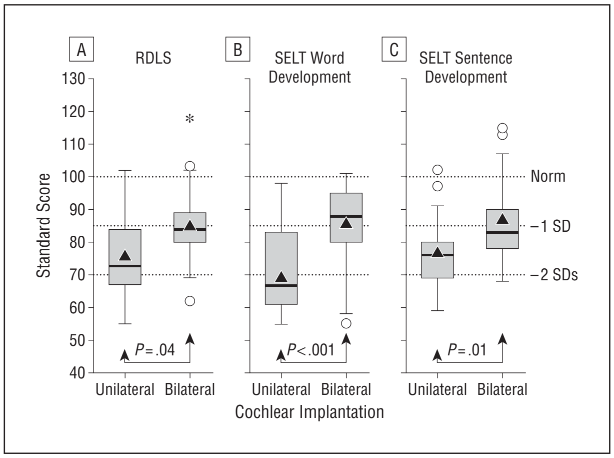

Figure 2. Standard scores of the unilateral and bilateral implantation groups on 3 language tests. A, Reynell Developmental Language Scales (RDLS). B, Schlichting Expressive Language Test (SELT) Word Development Subscale. C, SELT Sentence Development Subscale. The box plots represent the smallest observation, lower quartile, median (bold line), mean (triangle), upper quartile, largest observation, mild outliers (>1.5 times the interquartile range) (circles), and extreme outliers (>3 times the interquartile range) (asterisk).The dotted horizontal lines represent an age-appropriate language level (norm; top line), a language delay of −1 SD compared with the norm (middle line), and a language delay of −2 SDs compared with the norm (bottom line).

Although the bilateral implantation group obtained significantly better RDLS scores, in general their language levels were not age appropriate. Most of these children (14 [56%]) had a delay of more than 1 SD compared with the norm. However, 1 child in the bilateral implantation group performed even 1 SD above the norm (standard score, 118). The results of both groups are illustrated in Figure 2A.

SPOKEN LANGUAGE: EXPRESSION

The bilateral group (mean score, 86.1) performed significantly better on the Word Development Subscale of the SELT than did the unilateral group (mean score, 70.4) (mean difference [95% CI], 15.7 [5.9-25.4]). In the unilateral group, 14 children (56%) had a language delay of more than 2 SDs compared with the norm, whereas 3 children (12%) in the bilateral group had a delay of more than 2 SDs. The child in the bilateral implantation group who scored markedly high on the RDLS also obtained a very high score on the SELT Word Development Subscale (standard score, 127). This child underwent simultaneous implantation at 1.02 years of age. In the bilateral group, the child with the lowest score on the RDLS (standard score, 62) also obtained the lowest score on the SELT Word Development Subscale (standard score, 55). This child underwent sequential implantation and had an additional learning disability. Figure 2B illustrates the outcome on this subscale.

On the Sentence Development Subscale of the SELT, the bilateral group (mean score, 86.8) performed significantly better than did the unilateral group (mean score, 77.0) (mean difference [95% CI], 9.7 [1.5-17.9]). In the unilateral group, 7 children (28%) had a language delay of more than 2 SDs compared with the norm, whereas 2 children (8%) in the bilateral group had a delay of more than 2 SDs. Although the bilateral implantation group achieved significantly better scores, in general their performance was not age appropriate on the Sentence Development Subscale. Most of the children (13 [52%]) had a delay of more than 1 SD compared with the norm. The outcomes of the 2 groups are illustrated in Figure 2C.

INTERVAL BETWEEN FIRST AND SECOND IMPLANTATION

First, within the bilateral group, we compared the 8 children who underwent simultaneous implantation with children who underwent sequential implantation. To match both subgroups on age at first fitting, children had to undergo implantation before 2.00 years of age. A group of 14 children undergoing sequential implantation (82%) (Seq in Table 1) met this criterion and did not differ significantly from the 8 children undergoing simultaneous implantation on age at first fitting (Mann-Whitney test, 32; $P=.10$) or additional disabilities (likelihood ratio, 1.1; *df*, 2; $P=.59$). One of these 14 children undergoing sequential implantation received the second implant within the first year after the first implant. Among the other 13 children, 8 received their second implant within the second year and 5 within the third year after the first implant. The children undergoing simultaneous implantation did not perform better on the RDLS (Mann-Whitney test, 47.5; $P=.57$) or the SELT Sentence Development Subtest (Mann-Whitney test, 28.0; $P=.06$) than the children who underwent sequential implantation. However, the children undergoing simultaneous implantation performed significantly better on the Word Development Subtest of the SELT (Mann-Whitney test, 21.0; $P=.04$).

Second, the effect of the interval between the first and second implantation was analyzed, including all 25 children in the bilateral implantation group. There was an equal distribution of children receiving their second implant within the first (9 children [36%]), the second (9 [36%]), and the third (7 [28%]) year after the first implant. The shorter the interval, the better the results were on the RDLS ($\beta=-0.36$ [SE=0.17]; $R=0.40$; $P=.04$) and the SELT Word Development Subtest ($\beta=-0.58$ [SE=0.21]; $R=0.50$; $P=.01$) and Sentence Development Subtest ($\beta=-0.54$ [SE=0.18]; $R=0.53$; $P=.006$). The intervals between implantations and standard scores are illustrated in **Figure 3**.

COMMENT

This study compared language test scores of children undergoing bilateral and unilateral implantation 3 years after the first implantation. On the receptive RDLS and the SELT subtests, the bilateral group achieved significantly higher scores than the unilateral group. In this study, great care was taken to control for several factors. Evidently, some factors remain unknown, such as the exact socioeconomic status of the family at the time of testing. Because our population does not allow a randomized controlled trial, a case-control retrospective design passes the highest possible level of evidence. The high

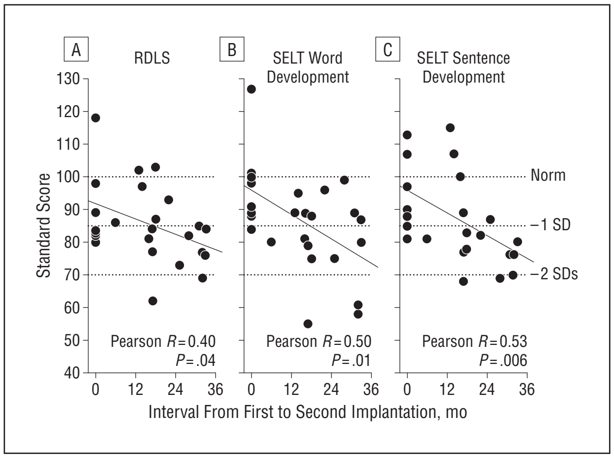

Figure 3. Standard scores in function of time from the first to second cochlear implantation. A, Reynell Developmental Language Scales (RDLS). B, Schlichting Expressive Language Test (SELT) Word Development Subscale. C, SELT Sentence Development Subscale. Each dot represents a child undergoing bilateral implantation, and the diagonal line represents the linear regression line. The dotted horizontal lines are described in the legend to Figure 2.

number of auditory, child, and environmental variables on which both groups were matched consolidates the hypothesis that the established difference in language scores is mainly attributed to bilateral implantation.

Twelve months after implantation, Tait et al[17] found a positive effect of bilateral implantation on the use of vocalization and audition to communicate. Their bilateral group was twice as likely to respond vocally to adults through audition alone. This indicated that the improved perceptual abilities, caused by the second cochlear implant, enabled more relaxed and vocally productive communication without any need to look at the adult. Improved speech recognition skills can facilitate the ability to pick up and learn language. This advantage of bilateral implantation evolves into the significantly higher language scores 3 years after implantation shown in the present study.

Although the primary goal of cochlear implantation in deaf children is to optimize their auditory abilities, a major final aim is to achieve good oral language skills. Strong language skills not only have a tremendous effect on communication but also affect a wide range of other aspects of life. For example, children with hearing loss often show higher levels of behavioral problems compared with hearing children. Once the language abilities of children with hearing loss are taken into account, the negative effect of hearing loss on behavior disappears.[18] In addition, strong language skills are essential to attend a regular school and achieve good academic results. In clinical treatment of deaf children receiving cochlear implants, factors related to language development should be considered seriously in combination with knowledge on costs, risks, and other long-term outcomes (eg, social and emotional development).

A shorter interval between the first and the second cochlear implant, and consequently more experience with the second cochlear implant, had a positive effect on the language results. Moreover, children undergoing simultaneous implantation achieved higher Word Development Subscale scores than did those undergoing sequential implantation. The inverse correlation of interval between implantations or experience with the second implant and speech recognition outcomes has been reported several times.[3,7,10,12] However, more research is needed to gain full insight into the effect of the interval between the first and second implantation on language development, especially with regard to long-term outcomes. The present study focused on short-term results. The effect of bilateral implantation and the interval between the first and second implantation may evolve over time.

In the present study, the positive effect of bilateral implantation on the RDLS and SELT results provides evidence of a benefit of bilateral implantation on semantic and syntactic language skills. However, the influence of bilateral implantation on other language aspects, such as morphologic characteristics and pragmatics, is still unknown. These language aspects might benefit even more from enhanced auditory skills through bilateral implantation. A more detailed evaluation of a broad range of language skills after a longer period of cochlear implant use will be performed in a subsequent prospective study.

CONCLUSIONS

The present results demonstrate a positive effect of bilateral implantation on expressive and receptive spoken language scores in prelingually deaf children 3 years after the first cochlear implantation. The interval between the first and second cochlear implantation was correlated negatively with all 3 language scores. An advantage of simultaneous compared with sequential implantation was noticed on short-term expressive Word Development scores. Although several studies have shown the positive effect of bilateral implantation in children on auditory development, our study is one of the first, to our knowledge, to demonstrate a positive effect on language test scores. Results from this study carry implications for the clinical treatment of deaf children receiving cochlear implants.

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Author Contributions: Ms Boons 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 analysis. *Study concept and design:* Boons, Brokx, Peeraer, and van Wieringen. *Acquisition of data:* Boons, Frijns, Philips, and Vermeulen. *Analysis and interpretation of data:* Boons, Peeraer, Wouters, and van Wieringen. *Drafting of the manuscript:* Boons and Brokx. *Critical revision of the manuscript for important intellectual content*: Boons, Frijns, Philips, Vermeulen, Wouters, and van Wieringen. *Statistical analysis:* Boons and Wouters. *Obtained funding:* Peeraer and Wouters. *Administrative, technical, and material support:* Boons, Frijns, Philips, and Wouters. *Study supervision:* Brokx, Frijns, Peeraer, Wouters, and van Wieringen.

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