

Long-Term Language Development in Children With Early Simultaneous Bilateral Cochlear Implants

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Objectives: This longitudinal study followed the language development of children who received the combination of early (5 to 18 months) and simultaneous bilateral cochlear implants (CIs) throughout the first 6 years after implantation. It examined the trajectories of their language development and identified factors associated with language outcomes.

Design: Participants were 21 Norwegian children who received bilateral CIs between the ages of 5 and 18 mo and 21 children with normal hearing (NH) who were matched to the children with CIs on age, sex, and maternal education. The language skills of these two groups were compared at 10 time points (3, 6, 9, 12, 18, 24, 36, 48, 60, and 72 months after implantation) using parent reports and standardized measures of general language skills, vocabulary, and grammar. In addition, assessments were made of the effects of age at CI activation, speech recognition abilities, and mothers' education on language outcomes 6 years after implantation.

Results: During the first 4 years after implantation, the gap in general expressive and receptive language abilities between children with CIs and children with NH gradually closed. While at the initial five to six assessments (3 to 36 months after implantation), significant differences between children with CIs and children with NH were observed; at 4 years after implantation, there were no longer any significant group differences in general language skills and most children with CIs achieved scores within 1 SD of the tests' normative means. From 2 to 3 years after implantation onward, expressive vocabulary and receptive grammar skills of children with CIs were similar to those of the reference group. However, from 4 years after implantation until the end of the observation period, 6 years after implantation, expressive grammar skills of children with CIs were lower than those of children with NH. In addition, a gap in receptive vocabulary appeared and grew increasingly larger from 4 to 6 years postimplantation. At the final assessment, the children with CIs had an average receptive vocabulary score around 1 SD below the normative mean. Regression analysis indicated that the children's language outcomes at 6 years after implantation were related to their speech recognition skills, age at CI activation, and maternal education.

Conclusions: In the first 4 years after implantation, the language performance of children with CIs became increasingly similar to that of their NH peers. However, between 4 and 6 years after implantation, there were indications of challenges with certain aspects of language, specifically receptive vocabulary and expressive grammar. Because these challenges first appeared after the 4-year assessment, the findings underline the importance of long-term language intervention to increase the chances of a continued language development comparable to that of NH peers. They also indicate that there is a need for comprehensive

longitudinal studies of the language development of children with CIs beyond 4 years after implantation.

Key words: Bilateral implantation, Child, Cochlear implant, Language, Language development.

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INTRODUCTION

Cochlear implants (CIs) have become a common means to allow children with congenital profound deafness to access sound and develop spoken language. Especially, children who receive CIs early in life can attain spoken language skills similar to those of their normal-hearing (NH) peers (Bruijnzeel et al. 2016; Colletti et al. 2012; Dettman et al. 2016; Geers & Nicholas 2013; Leigh et al. 2013; Niparko et al. 2010). Moreover, it has been shown that bilateral simultaneous implantation, as opposed to unilateral or sequential implantation, further facilitates access to sound, development of the brain's auditory pathways, and hence spoken language acquisition (Gordon et al. 2013; Kral et al. 2016). In this study, we followed the spoken language development of a group of children who were congenitally profoundly deaf and underwent both early (5 to 18 months of age) and simultaneous bilateral cochlear implantation. This was one of the first longitudinal studies of deaf children who have received this arguably best possible intervention, that is, children who have had bilateral access to their sound environment from very early in life. Our goal was to better understand the trajectory of these children's language development, the language outcomes that can be expected of them, and the factors that influence their language outcomes.

LANGUAGE DEVELOPMENT IN CHILDREN WITH EARLY COCHLEAR IMPLANTATION

The research literature strongly suggests that the best oral language outcomes are obtained when CIs are received at a young age (e.g., Leigh et al. 2013; May-Mederake 2012; Niparko et al. 2010; Quittner et al. 2016; Vandewalle et al. 2012). Specifically, implantation within the first year of life is associated with age-appropriate general language skills, whereas comparatively poorer outcomes have been observed for children who received implants later (Ching et al. 2009; Colletti et al. 2011; Holman et al. 2013; Tajudeen et al. 2010; Tobey et al. 2013). A recent meta-analysis by Ruben (2018) has examined 21 studies of language outcomes in children with CIs, with follow-up periods ranging from 3 years to >10 years. They concluded that CIs are most efficient in supporting language development when the child receives implants before the age of 12 months. After that age, the outcomes decline as the age of implantation increases. Notably, Colletti et al. (2012) have found evidence for an additional benefit of even earlier implantation, that is, before 6 months of age. Earlier age of implantation has also

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been associated with more rapid spoken language development, which means that children who receive CIs earlier have combined benefits of beginning to learn spoken language earlier and of learning it more quickly (e.g., Niparko et al. 2010; Tomblin et al. 2005). However, some studies have found that the advantage of early implantation for language development can diminish over time, suggesting that other variables may override the effect (Dunn et al. 2014). Colletti et al. argue that the long-term effect of age at implantation might be less apparent for general language comprehension, but it has a lasting effect on more specific and complex abilities within the domains of phonetics, grammar, and semantics because these abilities depend strongly on the functional specialization of certain networks in the brain that is triggered by sensory input during early sensitive periods of neuronal development. In a recent study, Nicholas and Geers (2018) have used spontaneous speech samples to assess the performance of children with CIs in the domains of expressive vocabulary, morphology, and syntax. The authors found that all three domains benefitted equally from early implantation.

As a result of the findings in this line of research, children who are congenitally deaf are receiving implants at an increasingly younger age, with many countries aiming for an implantation within the first 2 years of life (Bruijnzeel et al. 2017; Ramsden et al. 2012).

BENEFITS OF BILATERAL COCHLEAR IMPLANTATION

In addition to the increasingly earlier implantation, a growing number of deaf children now receives bilateral CIs, either sequentially or simultaneously (Peters et al. 2010; Ramsden et al. 2012). Bilateral implantation allows CI users to take advantage of bilateral auditory cues, which can facilitate listening in complex auditory environments by improving spatial hearing abilities such as sound localization (Asp et al. 2012; Grieco-Calub & Litovsky 2010; Litovsky et al. 2006; Yi et al. 2015) and the segregation of speech from background noise (Galvin et al. 2007; Litovsky et al. 2006; Van Deun et al. 2010). The improved access to sound in noisy listening situations is especially valuable for young children, who spend much of their time learning and socializing in acoustically challenging environments (Busch et al. 2017; Manlove et al. 2001; Shield et al. 2015). In such situations, bilateral stimulation likely helps to reduce listening effort (Hughes & Galvin 2013) and facilitates incidental learning (Sarant et al. 2014). This presumed benefit of bilateral implantation is supported by a range of studies showing that children with bilateral CIs achieve better language outcomes than children with unilateral CIs do (Boons et al. 2012b, 2013; De Raeve et al. 2015; Jacobs et al. 2016; Lammers et al. 2014; Leigh et al. 2013; Sarant et al. 2015; Sarant et al. 2014). For example, Boons et al. (2012b) found that the children who received bilateral CIs achieved significantly better spoken language expression and comprehension scores than did a matched group of children with unilateral CIs, even though both groups had received their CIs by 2 years of age. Moreover, the shorter the interval between the two implantations, the better the children's language performance. Similarly, Guo et al. (2015) found that at 1 year after implantation, children with bilateral CIs, but not children with unilateral CIs, demonstrate sensitivity to statistical characteristics of words comparable to that of children with NH during the early stages of lexical development (for

a review on statistical learning abilities of CI users, see Deocampo et al. 2018). There is also evidence suggesting that children who receive simultaneous bilateral implantation perform better than their unilaterally implanted peers do on measures of verbal reasoning (Jacobs et al. 2016).

For the maturation of the brain's auditory system, early access to hearing is critical and a lack of auditory stimulation can cause deviations in neural development that have long-lasting effects on auditory development, language acquisition, and even higher-level cognitive abilities (Kral et al. 2016; Kronenberger et al. 2014; Sharma & Campbell 2011; Werker & Hensch 2015). Similarly, unilateral auditory input can cause asymmetrical development, which can compromise the way that the auditory system responds to stimulation from a subsequent implant in the contralateral ear (Gordon et al. 2013; Jiwani et al. 2016; Litovsky et al. 2006). Notably, it has also been argued that, although early simultaneous bilateral implantation promotes symmetrical development of the auditory pathways more than unilateral or sequential implantation does, the cortical processing of sounds will still not be normal (Easwar et al. 2017).

WHAT CAN WE EXPECT OF CHILDREN WHO RECEIVE EARLY SIMULTANEOUS BILATERAL CIS?

In summary, better language outcomes can be expected for children who receive implants early and for children who receive bilateral implants. Therefore, it seems likely that the best intervention to promote oral language development for children who are profoundly deaf is early simultaneous bilateral cochlear implantation. With the exception of Jacobs et al. (2016), most studies of children with bilateral implants have not focused on the benefits of simultaneous implantation. Consequently, little is known about the language trajectories of children who have received simultaneous bilateral CIs at an early age. The decision whether bilateral cochlear implantation is done sequentially or simultaneously is affected by numerous factors and varies by clinic and country (Uecker et al. 2019). In Norway, simultaneous bilateral implantation has been the standard treatment since 2004 for children with profound deafness (Wie 2010). The first cohort of Norwegian children who have received simultaneous bilateral CIs has been followed longitudinally, and in the present study, we report results regarding their language development throughout their first 6 years after implantation.

PRESENT STUDY

We prospectively followed a group of 19 children who were congenitally profoundly deaf and who had received simultaneous bilateral CIs at 5 to 18 months of age. The purpose of this investigation was to understand what language outcomes deaf children can achieve if they receive this arguably optimal treatment. Therefore, we repeatedly assessed their language abilities throughout the first 6 years after the implantation and compared their performance to that of a matched cohort of children with NH who underwent the same testing protocol. The longitudinal design allowed us to follow the development of individual children from a very young age and to closely observe the development of their language abilities. It also allowed us to determine whether variation in background characteristics can predict language outcomes. We hypothesized that our sample of children, who have

received a combination of early and simultaneous bilateral cochlear implantation, had no additional deficits, were mainstreamed, and received spoken language stimulation, would gradually close the language gap existing between them and their NH peer.

MATERIALS AND METHODS

Participants

The study followed two groups of children: one group of children who were congenitally profoundly deaf and had received simultaneous bilateral CIs early in life and one matched reference group of children with NH. The first group initially included 21 children with CIs who were prelingually deaf (11 girls and 10 boys). All children were congenitally profoundly deaf except for one child who became deaf after contracting meningitis at the age of 4 months. All children but one received simultaneous implants in both ears; the exception was one child who had received the two implants 3 months apart. The mean age at implantation was 11.3 months ($SD = 3.86$; range: 5.5 to 18.9). The sample contained all children in Norway who had received bilateral CIs at 5 to 18 months of age between 2004 and 2007 and who had no known additional handicap. Nine Norwegian children with early simultaneous bilateral CIs were not included because they had severe additional handicaps and were unable to perform the language tests. For the 21 included children, neither parental reports nor medical records gave any indication of an additional handicap believed to affect hearing or language development. Furthermore, the parents of all children reported that the CIs were used throughout the day, except for one child who occasionally had some short periods of nonuse. Detailed demographic data of the children in the CI group are presented in Table 1.

The second group was a matched reference group. It consisted of 21 typically developing children with NH (pure-tone averages of <25 dB HL). These children were pairwise matched with the children in the CI group by age and sex. Maternal education was matched on the group level. They were recruited from healthcare centers, daycare facilities, and preschools in rural and urban communities in Norway. None of the children with NH had language delays according to reports from parents and educators. Two children with NH dropped out of the study after the 24- and 48-month assessments and were replaced in subsequent assessments by suitable matches.

Two of the CI users had to be excluded from the final data analysis, along with their NH matches, because they had periodic device failures, and consequently were stimulated unilaterally for over 9 months. Notably, one of the children that were included in the study had received a new implant (i.e., had a reimplantation) in one ear when 5 years of age but was kept in the study because they had been only 4 months without bilateral stimulation. Thus, the sample used for all further analysis consisted of 19 children.

All children, except for one in each group, had nonverbal intelligence within the normal range, that is, a standard score between 85 and 120 on Raven Colored Progressive Matrices (CPM; Raven 2004). The mean standard scores on this test were 101 for the CI group ($SD = 12.27$; range: 70 to 120) and 96.32 for the NH group ($SD = 10.39$; range: 70 to 115). An independent samples *t* test showed no significant difference between the two groups [$t(36) = 1.47$; $p = 0.150$; $d = 0.05$].

All but 2 children in each group had parents whose native language was Norwegian. In these families, both Norwegian and the

parents' native language were in use among family members, but the parents reported Norwegian to be the primary language for the child. Moreover, the children attended daycare centers and schools in which Norwegian was the main language. According to parent reports, all children with CIs predominantly used spoken language (auditory oral/verbal) and received professional language intervention after implantation. For most of the children, this intervention was some form of auditory-verbal therapy. In general, the intervention was most intensive in the first 24 months of life. Three of the children were in a special education learning environment, and 2 of these children had parents and teachers who sometimes used sign-supported spoken language to visually highlight the main words in their spoken language. All children were enrolled in a mainstream school setting from when they began formal education (i.e., August of the year the child turned 6).

Instruments

The data reported here include information from hospital records, parental questionnaires, and standardized tests that was recorded at different intervals after the implantation.

Questionnaires • Biographical information regarding cause of deafness, age of implantation, age at CI activation, type of implant, and duration of CI use was obtained from medical records. In addition, parents were asked to provide information regarding socioeconomic status (including the mother's education); the child's educational situation; the teachers', parents', and child's mode of communication; and the degree of support from hearing professionals given to caregivers and parents.

Parental assessment of the child's language development was obtained through a Norwegian translation of the Comprehension–Conceptual subscale from the Minnesota Child Development Inventory parent questionnaire (MCDI; Ireton et al. 1977). The Comprehension–Conceptual subscale includes measures of grammatical development and receptive vocabulary. Results from the MCDI were expressed as standardized scores with a normative mean of 100 and a standard deviation of 15. Scores between 85 and 115 define the normal range (mean ± 1 SD).

Nonverbal Intelligence • Raven CPM was used to test the children's nonverbal intelligence. Results were used as an indication of whether the child was within the normal range of intelligence.

Speech and Language Tests • Due to a lack of a single set of measures that could be used to longitudinally track performance of the children in this cohort, we selected the following set of tests which allowed evaluation of the children's speech recognition and language development at every age.

Speech Recognition • Speech recognition was measured by a phonetically balanced, single-syllable word-repetition test (Wie et al. 2007). The measure of interest on this test was the percentage of correctly repeated words.

Overall Receptive and Expressive Language • The children's overall language abilities were assessed by a Norwegian translation of the Mullen Scale of Early Learning (MSEL; Mullen 1995). The receptive (MSEL-RE) and expressive (MSEL-EX) subscales were used in the present study. The receptive language scale includes evaluation of general language knowledge, auditory comprehension, and auditory memory. The expressive language scale includes assessment of speaking ability, verbal analogies, vocabulary, and verbal memory tasks such as object naming and repetition of numbers and sentences. Results

TABLE 1. Demographic information for children with CIs in the original sample (n = 21)

Sex	Etiology	Age at (mo)			CI Model			Speech recognition (%)*
		Diagnosis	Hearing aid	CI implantation	Left	Right	Reimplantations	
Female	JLNS	0	—	5	Cochlear Nucleus	Cochlear Nucleus	—	96
Male	JLNS	1	2	5	MED-EL Pulsar	MED-EL Concerto	1 left	92
Male	Connexin 26	0	1	7	MED-EL Pulsar	MED-EL Concerto	—	91
Female	JLNS	0	1	8	Cochlear Nucleus	Cochlear Nucleus	—	94
Male	Unknown	4	6	8	Cochlear Nucleus	Cochlear Nucleus	—	96
Female	CMV	5	5	8	Cochlear Nucleus	Cochlear Nucleus	—	92
Female	Connexin 26	1	4	8	MED-EL Pulsar	MED-EL Concerto	—	96
Female	DFNB4	2	2	8	MED-EL Pulsar	MED-EL Concerto	2 right	76†
Male	Birth damage	1	2	9	Cochlear Nucleus	Cochlear Nucleus	1 right	86†
Female	Connexin 26	6	7	10	Cochlear Nucleus	Cochlear Nucleus	—	96
Male	Connexin 26	3	3	10	Cochlear Nucleus	Cochlear Nucleus	—	92
Female	Connexin 26	3	5	11	MED-EL C40+	MED-EL C40+	—	96
Female	Connexin 26	4	4	11	MED-EL Sonata	MED-EL Sonata	—	90
Male	Unknown	2	3	12	MED-EL Pulsar	MED-EL Concerto	—	84
Female	Waardenburg	2	9	12	MED-EL Pulsar	MED-EL Concerto	—	88
Male	Meningitis	9	—	13	MED-EL Pulsar	MED-EL Concerto	—	84
Female	DFNB4	8	—	14	MED-EL Sonata	MED-EL Sonata	—	88
Male	CMV	11	12	16	MED-EL Sonata	MED-EL Sonata	—	96
Male	Unknown	11	13	16	MED-EL Sonata	MED-EL Sonata	—	90
Female	Unknown	13	—	17	MED-EL Pulsar	MED-EL Concerto	—	86
Male	Unknown	3	3	18	MED-EL Pulsar	MED-EL Concerto	—	92
M		4.24	4.82	10.76				90.52
SD		3.96	3.59	3.79				5.25

All Cochlear Nucleus users had Cochlear Nucleus 24RE(CA).

*Best score obtained during all test moments.

†These 2 children were excluded from the analysis because they had more than extended periods (>9 mos) of unilateral stimulation due to issues with the CI.

CI, cochlear implant; CMV, Cytomegalovirus infection; DFNB4, deafness, autosomal recessive 4, with enlarged vestibular aqueduct (indicates Pendred-related syndrome), JLNS, Jervell and Lange-Nielsen syndrome; M, mean.

from the MSEL are expressed in *t* scores. The *t* scores have a normative mean of 50 and a standard deviation of 10. Thus, *t* scores between 40 and 60 define the normative range.

Receptive Vocabulary • We used the British Picture Vocabulary Scale, second edition (BPVS II; Dunn et al. 1997) to assess the children's receptive vocabulary. The results are expressed in standard scores (mean = 100; SD = 15).

Expressive Vocabulary • Expressive vocabulary was measured with the picture-naming subtest of the Wechsler Preschool and the Primary Scale of Intelligence, third edition (WPPSI-III; Wechsler 2002). The scaled scores with 10 as the normative mean were transformed into standard scores (mean = 100; SD = 15).

Receptive and Expressive Grammar • Grammatical abilities were tested using the Test for Reception of Grammar, second edition (TROG-2; Bishop 2003) and the Grammatical Closure subtest of the Illinois Test of Psycholinguistic Abilities (ITPA; Kirk et al. 1967). TROG-2 measures receptive comprehension of grammatical constructions in language. Results are presented in standard scores (mean = 100; SD = 15). The Grammatical Closure subtest of the ITPA measures expressive grammatical skills such as the ability to mark singular-to-plural and possessive transformations. Scores are presented as scaled scores, with mean = 36 and SD = 6 in the norm population.

Procedure

The study had a prospective, longitudinal, matched-group design. Tests were administered 3, 6, 9, 12, 18, 24, 36, 48, 60,

and 72 months after implantation. A different selection of tests was conducted at different test moments: Nonverbal intelligence was tested with Raven CPM once between 5 and 6 years of age. General receptive and expressive language abilities were tested with the MCDI and MSEL at all test moments from 3 to 48 months after implantation. Speech recognition, receptive vocabulary (BPVS II), and expressive vocabulary (WPPSI-III) were tested from 24 to 72 months after implantation. Receptive grammar (TROG-2) was assessed from 36 to 72 months, and expressive grammar (ITPA) from 48 to 72 months after implantation. Parent questionnaires and speech recognition tests were administered at all test moments.

The CI users were tested in a clinical setting, whereas the children with NH were tested at their daycare centers or at home with their parents. Qualified special education teachers administered all assessments. The CI users and the NH children were pairwise matched based on chronologic age at the point of each checkup. Independent samples *t* tests showed no significant difference in age between the two groups at any of the 10 assessments.

The number of children tested at the different assessment moments varied. Missing test results were due to missed appointments, or because a child, for various reasons, could or would not be tested. In addition, the oldest children in the sample were enrolled in the study only at the 24- or 36-month test moments.

The matched reference group of Norwegian children with NH compensated for the lack of Norwegian norms on some of the tests. Norwegian norms existed for TROG-2, BPVS II

(Lyster 2007), and WPPSI-III. The remaining language test results (MCDI, MSEL, ITPA) were interpreted according to U.S. norms. Due to the limited number of children in the Norwegian norming sample, age norms were available for only some age groups. If a child's raw score was lower than the lowest raw score for which there was a corresponding standard score, we used the lowest available standard score instead (left censoring).

The study was approved by the Regional Committees for Medical Research Ethics and the Data Inspectorate. All parents gave informed written consent to participate in the study and the children consented verbally.

Analyses

The independent samples *t* test was used to compare the mean language scores between groups. Two-tailed tests of significance were used, and the level of significance was set to 0.05, without correction for multiple comparisons. Cohen *d* was calculated as an indicator of effect size.

Multiple regression analyses were used to identify factors associated with the CI users' performance on the seven language tests at the study's endpoint, that is, the last moment when the test was used (48 or 72 months after implantation). As independent variables, we used maternal education, age at CI activation, and average speech recognition score across all test moments. Maternal education was assessed at the test moment and entered as an ordinal variable with five levels, ranging from elementary school education to more than 4 years of university education.

RESULTS

Speech Recognition

The average of the CI users' average speech recognition scores throughout the 10 test moments was 84% (SD = 4.58). The average for the matched group of children with NH was 93% (SD = 6.17), which was significantly higher than the mean of the CI group [$t(36) = -4.73$; $p < 0.001$; $d = 1.66$]. Table 1 shows the highest speech recognition scores that each child in the CI group had obtained during the assessments at 24 to 72 months of age.

Long-Term Language Development in Children With CIs Versus NH

All children with CIs were assessed on a battery of language tests at 10 measurement points throughout 6 years after they received their CIs (at 3, 6, 9, 12, 18, 24, 36, 48, 60, and 72 months postimplantation), and their results were compared with the scores of the NH matched reference group. The gap between the two groups evolved as a function of time since implantation and language test (for details, see Table 2).

General Language Development • From 3 to 48 months postimplantation, we obtained parent reports of the children's receptive and expressive language abilities using the MCDI and MSEL. The MCDI revealed differences between children with CIs and children with NH in the first 3 years after implantation (Table 2). The average scores in the CI group were typically lower than were the average scores in the NH group, showing a significant difference at the 12-month [$t(25) = 3.14$; $p = 0.004$; $d = 1.21$], 18-month [$t(28) = 3.69$; $p = 0.001$; $d = 1.35$], and 24-month intervals [$t(34) = 3.80$; $p = 0.001$; $d = 1.26$]. However,

at 36 and 48 months postimplantation, parents of children from the CI and NH groups rated their children's language abilities similarly, with no significant differences between the groups.

The children with CIs had receptive language scores on the MSEL-RE that were on average >1 SD below the normative mean and significantly lower than those of the matched NH reference group at the 3-month [$t(15) = 4.97$; $p < 0.001$; $d = 2.56$], 6-month [$t(17) = 2.74$; $p = 0.014$; $d = 1.27$], and 9-month intervals [$t(17) = 3.27$; $p = 0.004$; $d = 1.36$]. The CI users also scored significantly lower than did the NH group at the 12-month [$t(26) = -3.97$; $p < 0.001$; $d = 1.49$], 18-month [$t(28) = 2.98$; $p = 0.006$; $d = 1.09$], and 24-month testing intervals [$t(34) = 2.25$; $p = 0.031$; $d = 0.73$]. However, similar to the results of the MCDI, the difference in scores decreased with increasing CI experience, and at 36 and 48 months after implantation, there were no significant differences between the groups on the MSEL-RE. It should also be noted that at all assessments between 12 and 48 months after the implantation, mean scores of CI users were within 1 SD of the mean score of the test norms (Fig. 1).

The children's expressive language scores on the MSEL-EX showed that the CI group had significantly lower scores than did the NH reference group at the 3-month [$t(15) = -5.40$; $p < 0.001$; $d = 2.51$], 9-month [$t(21) = -2.25$; $p = 0.035$; $d = 0.94$], 12-month [$t(26) = -4.43$; $p < 0.001$; $d = 1.67$], 18-month [$t(28) = -3.75$; $p < 0.001$; $d = 1.37$], 24-month [$t(34) = -3.29$; $p = 0.002$; $d = 1.90$], and 36-month testing intervals [$t(36) = -2.30$; $p = 0.027$; $d = 0.75$]. At the 6- and 48-month assessment intervals, the differences between the two groups were not statistically significant. Notably, at 18 to 48 months postimplantation, the average score of children with CIs was within the normal variation (i.e., <1 SD below the normal mean).

Vocabulary Development • Receptive vocabulary knowledge was assessed yearly from 24 to 72 months postimplantation, using the BPVS II. At the 24- and 36-month testing intervals, there were no statistically significant differences in standard scores between the children with CIs and children with NH. However, the difference between groups was significant at 48 months [$t(36) = 2.43$; $p = 0.02$; $d = 0.88$], 60 months [$t(36) = 4.71$; $p < 0.001$; $d = 1.53$], and 72 months [$t(36) = 4.21$; $p < 0.001$; $d = 1.36$].

The CI group's average standard scores on the first four assessments were within 1 SD of the normative mean, but on the last assessment, 72 months postimplantation, the average standard score of the CI group was >1 SD below the normative mean (Fig. 1).

The children's expressive vocabulary was measured using the WPPSI-III picture-naming subscale at 24 to 72 months postimplantation. At none of the assessments were there significant differences between the WPPSI-III scores of children with CIs and children with NH. Moreover, the average scores for both the CI and the NH groups were within 1 SD of the normative mean at all assessments (Fig. 1).

Development of Grammatical Abilities • The difference between the groups on the measure of receptive grammar (TROG-2) was significant at the 48-month assessment [$t(35) = -2.21$; $p = 0.034$; $d = 0.72$], but not at 36 months [$t(19) = 0.02$; $p = 0.98$; $d = 0.01$], 60 months [$t(34) = 0.17$; $p = 0.86$; $d = 0.05$], or 72 months postimplantation [$t(35) = -0.88$; $p = 0.38$; $d = 0.05$]. On the test of expressive grammar (ITPA), there were significant differences between the two groups at 48 months [$t(31) = -2.62$; $p = 0.013$; $d = 0.88$] and 72 months

TABLE 2. Average language test results of children with CIs and the matched NH reference group 3-72 months after implantation

		Months after implantation																			
		3		6		9		12		18		24		36		48		60		72	
Group		CI	NH	CI	NH	CI	NH	CI	NH	CI	NH	CI	NH	CI	NH	CI	NH	CI	NH	CI	NH
Age at test moment																					
M		14.8	14.5	17.80	19.2	21.53	21.6	24.89	24.2	31.0	30.5	36.5	36.5	48.5	48.3	60.1	60.1	72.3	72.7	84.2	84.8
SD		3.4	3.5	3.71	3.2	4.24	3.3	4.47	3.7	4.2	4.1	3.8	4.0	4.2	4.5	4.3	4.2	4.3	4.0	3.9	3.7
General language (MCDI)																					
M		81.1	104.5	82.70	93.0	78.3	89.5	78.8	94.1	** 78.2	97.6	*** 79.6	100.8	*** 86.4	94.1	91.1	91.6				
SD		18.9	20.7	12.15	15.0	13.7	13.2	12.4	13.0	14.4	14.4	16.0	17.6	15.3	14.6	17.0	16.3				
n		11	4	10	6	13	11	14	13	16	14	18	18	19	17	18	18				
General language (MSEL-RE)																					
M		27.5	49.2	*** 37.10	47.7	*	36.6	53.5	** 41.9	58.5	*** 47.3	58.00	** 44.7	52.2	*	47.6	52.6	47.4	48.6		
SD		8.8	8.2	9.80	6.5	14.0	10.6	12.0	10.2	12.1	6.70	10.7	9.9	11.5	6.8	9.2	7.0				
n		11	6	10	9	11	12	14	14	15	15	18	18	19	19	19	19				
General language (MSEL-EX)																					
M		25.8	49.0	*** 40.6	44.6	39.5	45.6	*	38.4	48.7	*** 42.4	54.5	*** 42.7	54.1	** 43.0	51.6	*	47.2	53.2		
SD		6.7	11.2	6.6	2.9	6.4	6.6	6.4	5.9	10.3	7.1	12.6	7.5	13.9	8.4	11.9	6.5				
n		11	6	10	9	11	12	14	14	15	15	18	18	19	19	19	19				
Receptive vocabulary (BPVS II)																					
M																					
SD																					
n																					
Expressive vocabulary (WPPSI-III)																					
M																					
SD																					
n																					
Receptive grammar (TROG-2)																					
M																					
SD																					
n																					
Expressive Grammar (ITPA)																					
M																					
SD																					
n																					

Group means that are significantly different (independent samples t test, $p < 0.05$) are shown in bold. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$. BPVS II, British Picture Vocabulary Scale, standard score (M = 100; SD = 15); CI, cochlear implant; ITPA, Illinois Test of Psycholinguistic Abilities, standard score (M = 36; SD = 6) on the subscale Grammatical Closure; M, mean; MCDI, Minnesota Child Development Inventory, comprehension conventional subscale, standard score (M = 100; SD = 15); MSEL, Mullen Scales of Early Learning, standard score (M = 50; SD = 10) on the subscales receptive (MSEL-RE) and expressive language (MSEL-EX); n, number of participants; NH, normal hearing; TROG-2, Test for Reception of Grammar, second edition, standard score (M = 100; SD = 15); WPPSI-III, Wechsler Preschool and Primary Scale of Intelligence, third edition, standard score (M = 100; SD = 15) on the subscale Picture naming.

Children with CI versus Children with NH

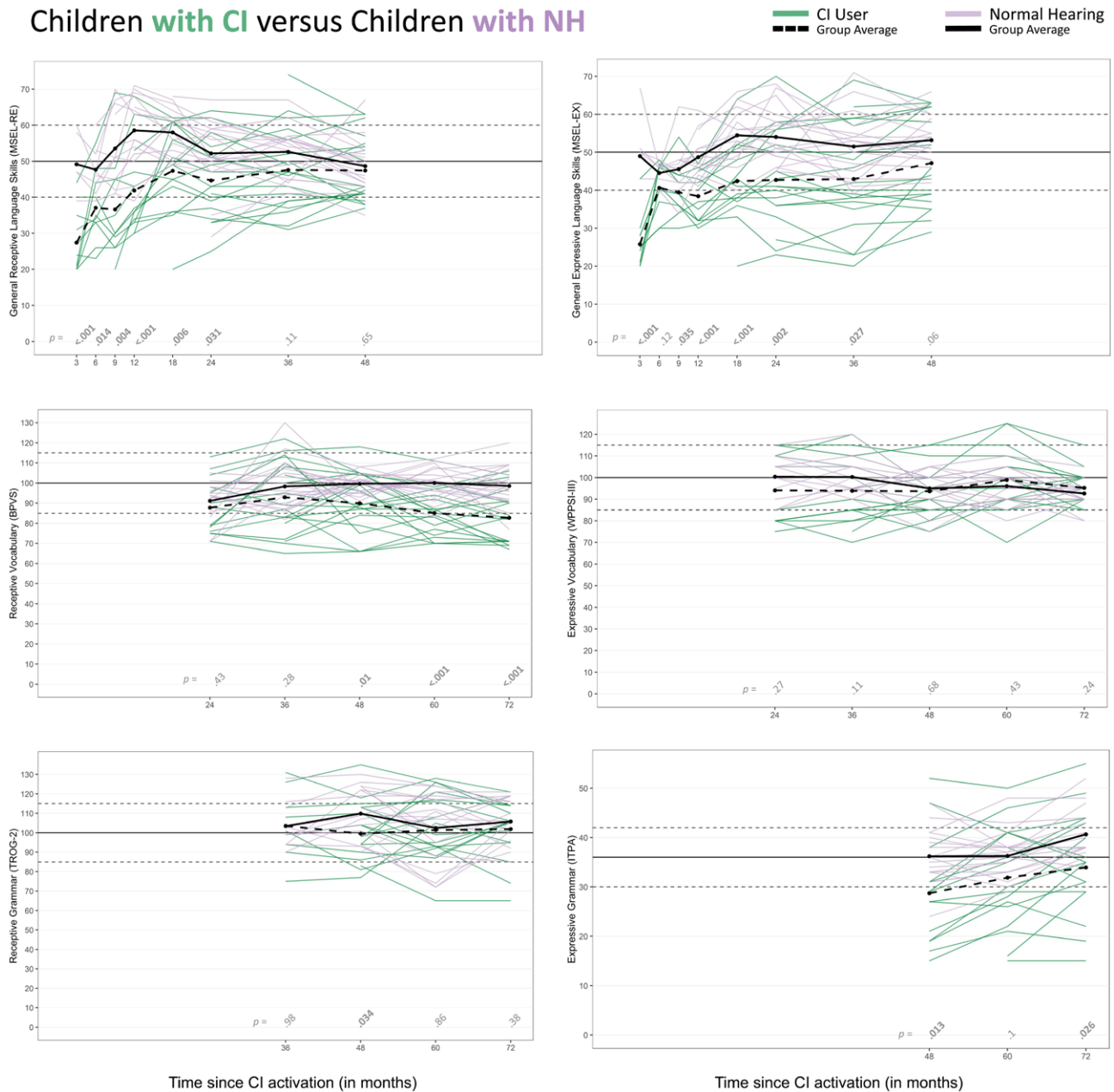


Fig. 1. Language performance of children with CIs and of children with normal hearing, using different receptive and expressive language measures in the first 6 yrs after implantation. The solid and dashed horizontal lines in the background show the normative mean \pm 1 SD. The significance levels (independent samples *t* tests) of the differences between the group means at all assessment moments are shown along the bottom of each panel. BPVS indicates British Picture Vocabulary Scale; CI, cochlear implant; ITPA, Illinois Test of Psycholinguistic Abilities; MSEL, Mullen Scale of Early Learning; TROG-2, Test for Reception of Grammar, second edition; WPPSI-III, Wechsler Preschool and the Primary Scale of Intelligence, third edition.

after implantation [$t(33) = -2.34; p = 0.026; d = 0.80$]. At 48 months, the CI group scored on average >1 SD below the normative mean on the test of expressive grammar (ITPA). On all other assessments of expressive and receptive grammar, both groups scored within 1 SD of the normative mean (Fig. 1).

Early Versus Later Implantation

There were 11 children (7 girls and 4 boys) who had received implants in the first year of life (mean = 8.79 months; SD = 2.00) and 8 children (3 girls and 5 boys) who received implants

during the age span 12 to 18 months (mean = 15.39 months; SD = 2.25). A comparison between the two groups using independent samples *t* tests showed that children who had received implants earlier obtained on average higher language scores on all 10 assessments than did the group of children who received implants later (Fig. 2).

Factors Associated With Endpoint Language Measures

Regression analyses with stepwise backward elimination of predictor variables showed that 48 months after implantation, the children's speech recognition ability explained 22% of the

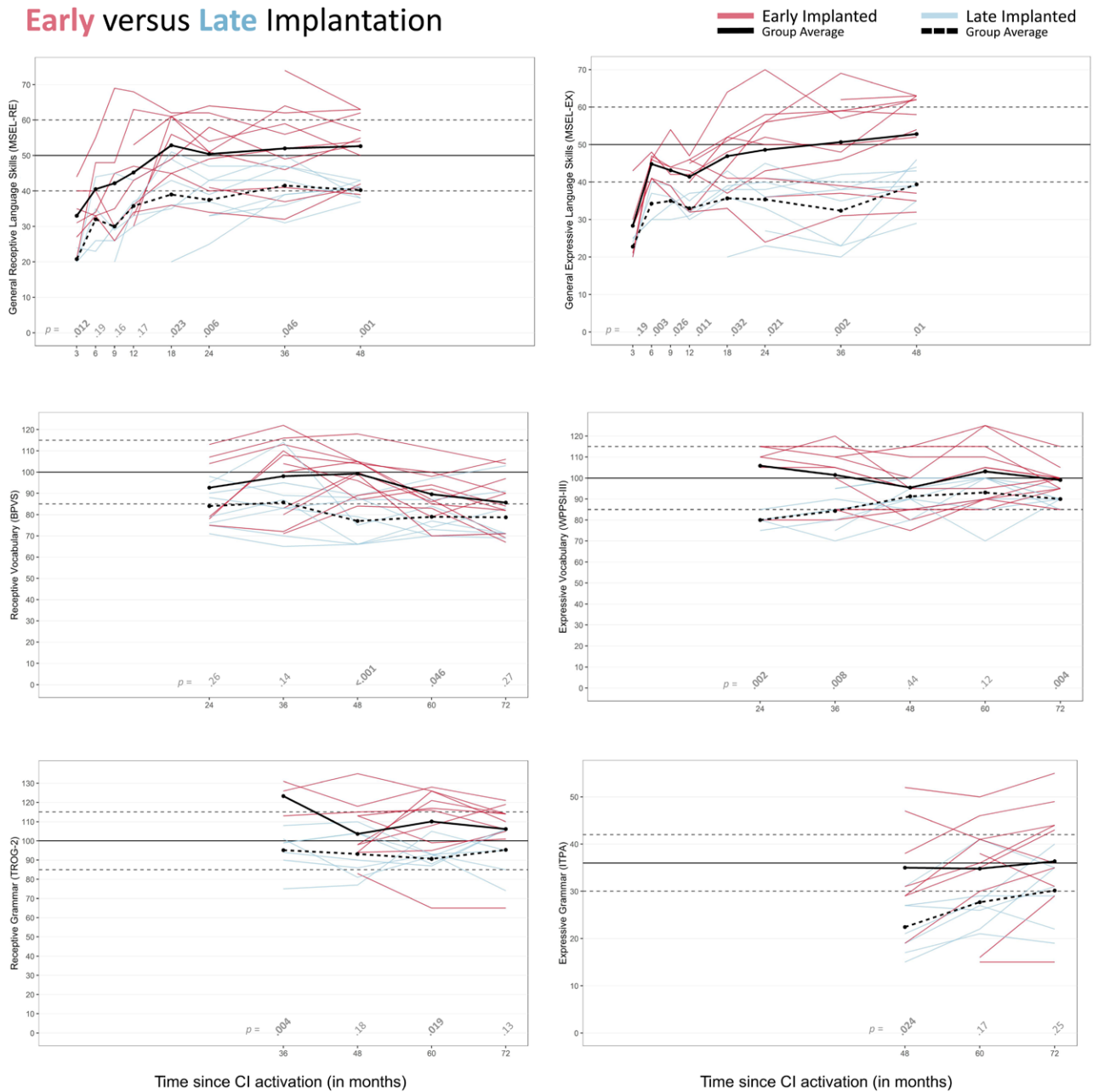


Fig. 2. Language performance of children who received CIs early (within 12 mos after birth) and of children who received CIs later (at 12 to 18 mos of age), using different receptive and expressive language measures in the first 6 yrs after implantation. The solid and dashed horizontal lines in the background show the normative mean \pm 1 SD. The significance levels (independent samples *t* tests) of the differences between the group means at all assessment moments are shown along the bottom of each panel. BPVS indicates British Picture Vocabulary Scale; CI, cochlear implant; ITPA, Illinois Test of Psycholinguistic Abilities; MSEL, Mullen Scale of Early Learning; TROG-2, Test for Reception of Grammar, second edition; WPPSI-III, Wechsler Preschool and the Primary Scale of Intelligence, third edition.

variation in the children’s general language abilities (MCDI) and 19% of the variation in general expressive language skills (MSEL-EX). Speech recognition ability and age at CI activation together explained 39% of the variation in general receptive language skills (MSEL-RE). At the assessment 72 months after implantation, maternal education was found to be an important predictor. In different combinations with speech recognition ability and age at CI activation, it explained between 37% (expressive vocabulary) and 75% (receptive grammar) of the

variation. In general, the predictors explained more variance in receptive than in expressive measures (see Table 3 for details).

DISCUSSION

In the present study, we prospectively followed the language development of children who were congenitally deaf, had received simultaneous bilateral CIs at an early age (before 18 months), and had no additional disabilities. Early simultaneous

TABLE 3. Results from multiple regression analysis, using background variables to predict cochlear implant users' standard scores on the language tests 48 or 72 months after implantation

Test	Months after implantation	Predictor	B	SE B	β	<i>t</i>	<i>p</i>	Adj. R ²
General language (MCDI)	48	Speech recognition (avg.)	1.96	0.81	0.51	2.43	0.027	0.224
General receptive language (MSEL-RE)	48	Speech recognition (avg.)	0.83	0.37	0.41	.276	0.041	0.389
		Age of CI activation	-1.16	0.44	0.49	2.64	0.018	
General expressive language (MSEL-EX)	48	Speech recognition (avg.)	1.25	0.55	0.48	2.26	0.037	0.186
Receptive vocabulary (BPVS II)	72	Speech recognition (avg.)	1.54	0.50	0.54	3.07	0.007	0.485
		Maternal education	3.43	1.57	0.38	2.18	0.044	
Expressive vocabulary (WPPSI-III)	72	Age of CI activation	-1.20	0.35	-0.54	-2.88	0.011	0.373
		Maternal education	2.06	0.94	-0.54	-2.88	0.011	
Receptive grammar (TROG-2)	72	Age of CI activation	-1.18	0.48	0.32	-2.49	0.025	0.752
		Maternal education	8.12	1.13	0.82	6.45	<0.001	
Expressive grammar (ITPA)	72	Maternal education	4.98	1.31	0.69	3.80	0.002	0.442

Predictor variables were selected using stepwise backward elimination.

Adj. R², adjusted multiple correlation coefficient; avg., average score of all test moments; BPVS II, British picture vocabulary scale; ITPA, Illinois Test of Psycholinguistic Abilities, Grammatical Closure subscale; MCDI, Minnesota Child Development Inventory; MSEL, Mullen Scales of Early Learning, receptive (MSEL-RE) and expressive language (MSEL-EX) subscales; SE B, SE of the regression coefficient; TROG-2, Test for Reception of Grammar, second edition; WPPSI-III, Wechsler Preschool and Primary Scale of Intelligence, third edition, picture-naming subscale.

bilateral implantation has arguably provided these children with the best possible starting conditions for spoken language development, and the goal of our study was to better understand the trajectory of their language development and the outcomes they can achieve. To that end, we compared their performance on different language assessments to that of a cohort of children with NH, matched for age, sex, and maternal education. The children's language abilities were repeatedly measured during the first 6 years after implantation, with 10 assessment points throughout this period. The consistent and frequent testing allowed us to examine the long-term trajectory of language development and to observe whether the children's language abilities catch up with those of their NH peers. The data also enabled us to study the impact of speech recognition abilities, maternal education, and age at CI activation on language outcomes.

Long-Term Language Development

We found that the gap between children who had received early simultaneous bilateral CIs and children with NH initially appeared to close. However, toward the end of the observation period, and when more specific aspects of language were tested, the gap reappeared. Specifically, most CI users showed a steady improvement of general language skills throughout the first 4 years after implantation (as measured with the MCDI and MSEL), and at 48 months after implantation, these tests did not show significant differences between the groups. However, from 48 to 72 months after implantation, and when the children were tested on specific aspects of language, they showed more mixed results. While they had age-appropriate expressive vocabulary, their receptive vocabulary increasingly fell behind that of the NH group, and at the final assessment, their mean score was >1 SD below the mean of the test's normative data. This result is in line with previous research in unilateral CI users, that has found slower vocabulary development compared with that of children with NH (Blamey et al. 2001; Connor et al. 2000; Le Normand et al. 2003; for a review, see Lund 2016). Because we did not have a unilateral CI group to compare with, we could not ascertain whether having bilateral CIs facilitated the vocabulary development of the children in our sample. However, compared with that of children with NH, vocabulary development was delayed. One explanation

for the initial closing and later reappearance of the vocabulary gap between children with CIs and those with NH could be that the words acquired at a later age are more difficult for children with CIs to learn, for example, because of increasing abstractness or lower usage frequency (Hansen 2017). The reappearing vocabulary gap might also reflect a change in the circumstances under which language is encountered: school-aged children spend more time in acoustically demanding environments, such as noisy classrooms (Busch et al. 2017), and more advanced vocabulary might more often be learned through overhearing conversations between others, which can be especially challenging for children with CIs (Boderé & Jaspaert 2017; Vermeulen et al. 2012).

The development of grammar showed similarly mixed results 36 to 72 months after implantation. On average, the receptive grammar of children with CIs was within 1 SD of the norm, but on the test of expressive grammar, they scored >1 SD below the norm and significantly lower than did the NH reference group 48 and 72 months after implantation. The difference between the groups corresponded to approximately 1 SD of the test's normative score. Difficulties in receptive and expressive grammatical development have also been reported previously, and it has been suggested that they reflect difficulties with the perception and production of morphologic markers that are less acoustically salient (Boons et al. 2013; Geers et al. 2009; Szagun 2004; Tomblin et al. 2015). It is conceivable that problems with the acquisition of grammatical markers surface relatively late because the incorrect use of these markers is common in typical language development at a younger age. For example, a cross-sectional study of 4- to 8-year-old Norwegian-speaking, typically developing children showed that performance on tests measuring past-tense inflections did not reach ceiling level until approximately 8 years of age (Ragnarsdóttir et al. 1999).

It is also possible that the language gap reappears toward the end of the observation period because literacy skills come into play at that time. Studies of typically developing children suggest that the development of literacy facilitates further language learning (Duff et al. 2015). While most children with CIs achieve reading skills within the average range for their age, a substantial minority of them experiences challenges in reading and writing development (for a review, see Mayer & Trezek

2018). For these children, poor literacy skills may represent an additional disadvantage for language development in the school years and onward.

Predictors of Language Outcomes

Multiple regression analysis showed that better speech recognition ability, earlier implantation, and higher maternal education were all predictive of better language skills 6 years after implantation. Correlations between speech recognition scores and language development have been shown before (Blamey et al. 2001; Desjardin et al. 2009; Eisenberg et al. 2016). They might indicate the benefits of better access to sound in difficult listening situations (Eisenberg et al. 2016) and of the improved ability to pick up acoustically nonsalient aspects of language—for example, short function words or subtle grammatical markers (Le Normand et al. 2003; Szagun 2004). However, it should be noted that the speech recognition test we used is itself language based and using it to predict language outcomes is—to some extent—circular, thus undermining causal interpretations.

The beneficial effect of early implantation has also been reported previously (Ching et al. 2009; Dettman et al. 2016). In our sample, we found a benefit of implantation before 12 months over implantation between 12 and 18 months of age. After 6 years of CI experience, most children who had received implants before 12 months of age obtained scores within the normal range for expressive vocabulary (100%), receptive grammar (91%), and expressive grammar (73%). Notably, only 46% of them had age-appropriate receptive vocabulary, while 2 years earlier, 91% had age-appropriate receptive vocabulary. Good receptive vocabulary performance 4 years after the implantation has also been found by Dettman et al. (2016), who reported that up to 81% of children who received CIs before the age of 12 months and 57% of children who received unilateral CIs between 13 and 18 months of age had attained scores on receptive vocabulary within the normal range after 4 years of CI experience. Our results seem to indicate that initially good vocabulary performance of children with CIs does not secure vocabulary development in line with that of NH children later on. Thus, long-term follow-ups may be necessary to get a full understanding of language development in children with hearing loss.

In our sample, at all 10 assessments, children who received implants between 12 and 18 months of age had lower scores than did those who had received implants earlier. By the end of the observation period, 6 years after implantation, they had not caught up. While it is conceivable that the persistent difference was an effect of the later age at implantation, there were other differences between the two subgroups that could explain some of the difference in performance. The children who had received CIs early were, for example, significantly more often deaf due to genetic causes, as opposed to infections (meningitis and cytomegalovirus) or unknown etiology (Fisher exact test, $p = 0.046$; see also Table 1). Our sample is too small to rule out such alternative explanations.

The effect of maternal education on language outcomes can be understood as a proxy for the effect of socioeconomic status. Socioeconomic status is well known to predict the trajectory of language development both in children with hearing impairments and in children with NH (e.g., Black et al. 2014; Hart & Risley 1995; Schjølberg et al. 2011; Zambrana et al. 2014). The influence of socioeconomic status might be mediated through aspects of parent–child communication (Hoff 2003; Rowe

2012) or through other differences in the environments of children from low- and high-income families (Evans 2004, 2006; Ferguson et al. 2013).

Implications for Clinical Practice

Previous studies have shown that early implantation alone is no guarantee for age-appropriate language development (Boons et al. 2012a; Geers et al. 2016; Niparko et al. 2010). This finding holds true for the children in the present study, who have received early simultaneous bilateral CIs. Thus, even under the most optimal circumstances in terms of age and mode of implantation, children with CIs are at risk for problems with language development. Our data also show that some of these problems might surface only long after the implantation: even when language performance at 48 mo post implantation is age appropriate, children with CIs might at a later point experience challenges in some areas of language. In our study, such challenges became apparent around the time that children entered school, 5 to 6 years after cochlear implantation. Thus, our findings indicate that children with CIs must be monitored closely and may require intensive and continuous intervention and support for many years after the implantation.

We found that some aspects of language are more likely to be at risk than others. From our results, it appears that receptive vocabulary and expressive grammar seem to be particularly challenging. Which aspects of the language of CI users are the most vulnerable and how exactly their development can be facilitated through intervention are questions for future research. For such research, it will be critical to develop and use assessment methods sensitive enough to identify specific weaknesses.

Strengths and Limitations

One strength of the present study was that we closely followed the children's language development over an extended period, namely at 10 assessment points throughout the first 6 years after cochlear implantation. This tight assessment schedule and the use of different language measures have allowed insights into the language development of children with CIs across different areas of language. Furthermore, we compared the children's performance with that of a matched reference group of children with NH, as well as with the tests' normative data. Another strength was that the sample of children with CIs was relatively homogeneous: all of them had received early simultaneous bilateral implantation, did not have additional disabilities thought to affect language and cognition, and were treated and followed by the same clinic.

A limitation of this study was the relatively small sample size, which reduced statistical power to detect differences between groups and to identify predictors of language development. We found a number of factors related to language outcomes, in particular, earlier implantation, better speech recognition ability, and higher maternal education. While these effects are in line with those found in previous research, larger samples are needed to determine which of them are generalizable beyond the relatively small cohort studied here.

To assess the robustness of our results, we performed a complementary analysis with mixed-effects models, in which we modeled the language performance taking into account the repeated measures and trying different strategies for handling missing data. This analysis has confirmed the results of the simpler

analyses reported here, which has increased our confidence that our results are robust, despite the relatively small sample size.

CONCLUSIONS

In the first 4 years after implantation, children who had received early simultaneous bilateral CIs showed gradually improving performance and an incremental closing of the gap to children with NH, both of which support the view that early and simultaneous bilateral implantation allow children with profound bilateral congenital deafness to achieve robust age-appropriate oral language skills and might be the best practice to support their oral language development (Ramsden et al. 2012). However, 4 to 6 years after implantation, we found a more mixed picture with respect to receptive vocabulary and expressive grammar. This mixed picture underscores the need for longitudinal investigations of language development that extend beyond 4 years postimplantation, the importance of assessing the language of children with CIs in detail, and the need to closely monitor and support these children for many years after the implantation.

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