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# Language development in children after receiving bilateral cochlear implants between 5 and 18 months

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#### ARTICLE INFO

# ABSTRACT

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Keywords: Cochlear implant Language development Expressive language development Receptive language development Prelingually deaf children Early bilateral implantation *Objective:* The objective of this study was to examine receptive and expressive language development in children who received simultaneous bilateral cochlear implants (CIs) between 5 and 18 months of age and to compare the results with language development in chronologically age-matched children with normal hearing.

*Methods:* The study used a prospective, longitudinal matched-group design. Data were collected in a clinical setting at postoperative cochlear implant check-ups after 3, 6, 9, 12, 18, 24, 36, and 48 months of implant use. The sample included 42 children: 21 cochlear implant users and 21 with normal hearing, matched pairwise according to gender and chronological age. Communication assessments included the LittlEARS questionnaire, the Mullen Scale of Early Learning, and the Minnesota Child Development Inventory.

*Results:* The cochlear implant users' hearing function according to LittlEARS was comparable to that of normal-hearing children within 9 months post-implantation. The mean scores after 9 and 12 months were 31 and 33, respectively in the prelingually deaf versus 31 and 34 in the normal-hearing children. The children's receptive and expressive language scores showed that after 12–48 months with cochlear implants, 81% had receptive language skills within the normative range and 57% had expressive language skills within the normative range. The number of children who scored within the normal range increased with increasing CI experience.

*Conclusions:* The present study showed that prelingually deaf children's ability to develop complex expressive and receptive spoken language after early bilateral implantation appears promising.

The majority of the children developed language skills at a faster pace than their hearing ages would suggest and over time achieved expressive and receptive language skills within the normative range. © 2010 Elsevier Ireland Ltd. All rights reserved.

# 1. Introduction

Language is an important tool for knowledge acquisition and participation in society throughout life. Delayed language development can have ripple effects on children's social life and education. Early, high-quality access to auditory linguistic stimulation is crucial for speech development. An absence of or reduced access to auditory stimulation has caused delayed or inadequate speech development in many hard-of-hearing children [1]. The implementation of neonatal hearing screening and the increased use of bilateral cochlear implants have provided more and more children with severe-to-profound deafness access to bilateral auditory signals throughout their first year of life.

The effect of early bilateral stimulation on prelingually deaf children's abilities to develop functional speech and language over time is still unexplored. It is recognized that early unilateral implantation is beneficial for early language competence [2,3]. However, it is unclear whether the age at which a profoundly deaf child receives two implants and the age at which the child attains normative mean language skills have long-lasting consequences for later linguistic competence. Studies by Hart and Risley [4] and Snow et al. [5] have shown that hearing experience in early childhood lays the foundation for more advanced use and acquisition of language. They also show that limited experience or, conversely, intensified stimulation leaves imprints that affect the direction and speed of later development. Spencer and Oleson [6] found that early access to unilateral cochlear implant input enables children to build better phonological processing skills. The early speech recognition and speech production skills gained from early CI input have been shown to predict children's reading skills. Hay-McCutcheon et al. [7] found that both the receptive and the expressive language ages in children with unilateral CI increased as the children with cochlear implants aged. However, the gap between the average performance for normal-hearing children and

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the overall mean performance in children with CI increased with chronological age.

Recent reports indicate that unilateral cochlear implantation in prelingually deaf children within the first year of life may result in speech and language skills comparable to those of children with normal hearing [2,8–11]. Previous studies of children with unilateral hearing loss, however, show evidence of difficulties with speech recognition in noisy environments, problems with locating sounds in their environments, and problems with the head-shadow effect [12]. A few recent studies show that bilateral cochlear implantation is important for auditory development and function [13–16]. Tait et al. [17] found that children with two CIs used significantly more audition and vocalization in communication after 12 months of CI use than unilaterally implanted children.

In prelingually deaf children, the implications of early bilateral implantation for spoken language development and ease of communication are of interest. Could the introduction of early bilateral cochlear implantation diminish the likelihood of delayed language development in prelingually deaf children and thus contribute to an improved foundation for educational and social development?

The aim of this study was to examine language development in prelingually deaf preschool children and their probability of developing and maintaining age-appropriate language over time. The intention was to explore to what extent prelingually deaf children who received bilateral simultaneous implantation in the fifth to eighteenth month of life develop age-appropriate language skills and how age of implantation affects children's language skills over time.

# 2. Materials and methods

# 2.1. Subjects

The study sample consisted of 21 profoundly prelingually deaf children (10 boys and 11 girls). The reference group consisted of 21 children with normal hearing (NH) (PTA < 25 dB HL). The CI users and the normal-hearing children were matched pairwise for gender and chronological age at the time of each examination. The two groups were matched groupwise based on mothers' education. The sample of prelingually deaf children included all the children in Norway who received simultaneous bilateral cochlear implantation between 5 and 18 months of age between 2004 and 2007 and who had no known additional disabilities assumed to affect language development. The reference group was randomly selected from health care centers and kindergartens in one rural and one urban community. Demographic and developmental features of the participants with CIs are given in Table 1. The average age of bilateral implantation was 11.3 months (SD = 3.9), and the range was 5.5-18.8 months. The average age of sound connection was 12.7 months (SD = 3.8), with a range of 7.2-19.9months. The average usage time was 37 months (SD = 10.4), with a range of 24-48 months. Fourteen children had Medel devices on both ears (models in use were the C40+ (one child), the PULSAR (eight children) and the SONATA (five children)). All used the continuous interleaved sampling (CIS) coding strategy in both implants. Seven children had Cochlear devices in both ears (Cochlear Nucleus 24RE (CA)) and used the advanced combination encoders (ACEs) coding strategy in both implants. The implants were used during all waking hours by all the children, with the exception of one child who had short periods during the day without the CI. All but two children in the CI group and two children in the reference group had parents whose native language was Norwegian. Four of the children were bilingual. An auditory oral/verbal habilitation approach was used for all the children. Three children were in a special education learning environment, and two children had parents and teachers who sometime used sign language to visually highlight the main words in their spoken sentences.

# 2.2. Assessment materials

Hearing and speech recognition were assessed via the LittlEARS parent questionnaire and single-syllable word tests. The LittlEARS questionnaire was designed to assess the development of auditory behavior in children during the first two (hearing) years [18]. Note that in the present study, the CI users and the normal-hearing children were matched by chronological age (not hearing age) at the point of testing. The sample was tested at 3, 6, 9, and 12 months post-implantation. The children's ages ranged from 7 to 29 months at these four testing points. Results from the LittlEARS were expressed in raw scores. The single-syllable word test consists of phonetically balanced lists of 25 and 50 words familiar to children from approximately three to four years of age. Twenty-five words from the single-syllable word test were administered via live voice for children under 4 years.

The children's receptive and expressive language was tested using the Mullen Scale of Early Learning (MSEL) [19] and the Minnesota Child Development Inventory parent questionnaire (MCDI) [20]. Two MSEL subscales were used: the receptive language scale and the expressive language scale. The receptive language scale assesses auditory comprehension and auditory memory skills and includes items of general knowledge. The expressive language scale measures speaking ability and language formation and includes naming objects as well as repeating numbers and sentences. Results from the MSEL were expressed in T-scores with a normative mean of 50 and a standard deviation of 10. T-scores between 40 and 60 defined the normative range. The MCDI parent questionnaire uses yes/no questions to assess child development. Three MCDI subtests were used in this study: Expressive Language, Comprehension-Conceptual, and Situation Comprehension. The Expressive Language subtest assesses the child's expressive vocabulary and syntax development. The Comprehension-Conceptual subtest assesses receptive vocabulary and syntax development. The Situation Comprehension subtest assesses the child's development of understanding of the world around him/her. In the Expressive Language subtest, the agediscriminating scale samples primarily the first 3 years [20]. The Situation Comprehension subtest has been found to have a high correlation with intellectual functioning [21]. Results from the final administration of the Situation Comprehension subtest was thus used as a proxy for intelligence testing, as an indication of whether the child was within the normal range of intelligence. 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 defined the normative range.

# 2.3. Study design and implementation

The study had a prospective, longitudinal, matched-group design. Data was collected in a clinical setting at postoperative CI-check-ups after 3, 6, 9, 12, 18, 24, 36 and 48 months of CI-use. Note again that the CI users and the normal-hearing children were matched pairwise based on chronological age at the point of each check-up. *T*-test shows no significant difference in age between the two groups at the eight check-up times. The number of children tested at the different check-up points varies. Missing test results were due to missed appointments, or because a child, for various reasons, could or would not be tested. All normal-hearing children were tested at the day-care center or at home with their parents. All tests were administrated in Norwegian by qualified special education teachers. On all tests, items increase in difficulty and the

Sex	Etiology	Onset of profound loss (months)	Age at completed diagnose (months)	Age at hearing aid (months)	Age at operation (months)	User time (months)	Number of check-ups	Educational setting	Main Communication approach	Single-syllable word recognition
Female	Connexin 26	0	3	3	11	48	2	Special	Auditory-oral	96%
Male	Unknown	0	3	3	17	48	5	Main stream	Auditory-oral	88%
Male	Meningitis	4	9	9	13	48	6	Home schooled	Auditory-oral	84%
Female	JLNS	0	0.1	1	8	48	8	Main stream	Auditory-oral	62%
Female	Unknown	0	13	Not fitted	18	48	8	Main stream	Auditory-verbal	72%
Female	JLNS	0	0.2	Not fitted	5	48	8	Special	Auditory-oral	78%
Male	JLNS	0	0.5	2	5	48	8	Main stream	Auditory-verbal	72%
Female	Connexin 26	0	1	4	8	48	8	Main stream	Auditory-verbal	88%
Male	Birth damage	0	1	1	9	36	7	Main stream	Auditory-oral	72%
Female	Pendred syndrome	0	2	2	8	36	7	Main stream	Auditory-verbal	52%
Male	Unknown	0	2	3	12	36	7	Main stream	Auditory-verbal	72%
Male	Connexin 26	0	0.2	1	7	36	7	Main stream	Auditory-verbal	91%
Male	Cytomegalus virus	0	11	11	16	36	7	Main stream	Auditory-verbal	88%
Female	Waardenburg	0	2	9	12	36	7	Main stream	Auditory-verbal	70%
Female	Connexin 26	0	6	7	10	36	7	Main stream	Auditory-verbal	100%
Male	Connexin 26	0	3	3	10	24	6	Main stream	Auditory-verbal	
Female	Cytomegalus virus	0	5	5	8	24	6	Main stream	Auditory-verbal	100%
Female	Pendred syndrome	0	8	Not fitted	14	24	6	Special	Auditory-oral	72%
Male	Unknown	0	11	12	16	24	6	Main stream	Auditory-verbal	76%
Female	Connexin 26	0	4	4	11	24	6	Main stream	Auditory-verbal	90%
Male	Unknown	0	4	6	8	24	6	Main stream	Auditory-verbal	86%
Mean			4	4.8	11.3	37.1	6.5			80.5%
Min			0	0.75	5.5	24.0	2.00			52
Max			13	13.0	18.8	48.0	8.00			100
SD			4	3.7	3.9	10.0	1.4			12.7

Table 1 Demographic data of the prelingually deaf participants (N=21).

JLNS: Jervell and Lange-Nielsen syndrome.

testing proceeded until a specified ceiling of incorrect responses was reached. The language test results were interpreted according to American norms. The reference group of normal-hearing Norwegian children compensated for the lack of Norwegian norms on these tests. All the parents signed consent forms, and the study was approved by the Data Inspectorate and by the Regional Committees for Medical Research Ethics (REK).

# 2.4. Analyses

Pearson's product moment correlation was used for simple correlations. The independent samples *T*-test were used to compare the mean receptive and expressive language within and between groups. Repeated-measure ANOVA was used to examine changes over time. Two-tailed tests of significance were used, and the level of significance was set to 0.05, without correction for multiple testing. Eta squared was used to estimate effect size.

# 3. Results

The results from the MCDI Situation Comprehension subtest indicated that all the children had cognitive skills within the normal range. There was no significant difference between the mean score calculated across all check-up points for the CI users (118) and the reference group (122) (t = -0.46, p = 0.65).

# 3.1. Listening performance

The results from the LittlEARS questionnaire showed that after 9 months of bilateral CI use, CI users had achieved an average score in listening performance equivalent to the scores of their peers with normal hearing. There was no significant difference between the two groups' average scores at the 9- and 12-month check-ups (mean score of 30 versus 31 and 33 versus 34, respectively) (z = -0.970, p = 0.33, z = 1.190, p = 0.23) (see Fig. 1). Among the CI users, the children who were the youngest at the time of sound connection had the highest scores. The correlation between age at connecting and obtained listening age quotient was significant at the 3- through 12-month check-ups (r = 0.58, p = 0.003) (r = 0.57, p = 0.005) (r = -0.44, p = 0.01) (r = 0.68, p = 0.00). The CI users' single-syllable word recognition test scores are presented in the last column in Table 1. These scores refer to the highest score obtained after 24-48 months user time. Scores from one child were not obtained due to young age and unwillingness to complete the



**Fig. 1.** LittlEARS raw scores at 3, 6, 9 and 12 months post-implantation attained by children with cochlear implant at age 7–29 months and their normal-hearing peers matched on gender and on chronological age at check-up.

test. The results showed that the CI users on average performed significantly lower than the reference group: 80.5 (SD = 12.7) versus 91.9 (SD = 6.5) (t = -3.5, df = 30, p = 0.00). The magnitude of the difference in the means was strong (eta squared = 0.29).

# 3.2. Receptive language

Results from the MSEL receptive language scale showed that CI users at the 3- through 9-month postoperative check-ups achieved scores below minus one standard deviation from the normative mean. However, after 12 months and through 48 months of use, the scores were within the normal range of one standard deviation from the normative mean (Fig. 2). The CI users' mean scores were lower than the scores of their normal-hearing peers, with the difference reaching statistical significance at the 3- through 24-month post-implantation check-ups (Table 2). The magnitude of the difference in the means was strong (eta squared = 0.62, 0.33, 0.35, 0.41, 0.27, 0.17, at the 3- through 24-month check-ups,



Fig. 2. Average receptive and expressive language performance over time for all 21 Cl users and their chronologically age-matched, normal-hearing peers, measured by the Mullen Scales of Early Learning. Results are expressed as *T*-scores. It should be noted that not all 42 subjects have data at each data point.

	Tests		3 month:		6 months		9 months		12 month	S	18 mont	hs	24 mont	hs	36 mont	hs	48 month	6
			C	HN	a	HN	CI	HN	CI	HN	CI	HN	CI	HN	CI	HN	CI	ΗN
Receptive	MSEL	Mean	27.5	49.2	36.5 0.5	47.3 6.7	36.3 1 2 A	53.0	39.9 17.4	57.9 0.8	44.2 1 / /	57.5 7.0	43.1 11 2	52.6 0.7	45.4 111	53.7 6.6	48.9 ° 7	49.2 6 4
тапуцаўс		n v	0.0 11	9.7	ر.و 11	0.2 10	12.12	14.0	12.4 16	э.о 16	14.4	17	20	20	14.4 15	0.0 14		6.0
		T-test	t = -5.0;	<i>p</i> = 0.00	t = -3.0;		t = -3.6;		t = -4.5;		t = -3.4;	<i>p</i> = 0.00	t = -2.9;	<i>p</i> = 0.00	t = -1.9;	<i>p</i> = 0.08	t = -0.7; p	= .95
	MCDI	Mean	81.1	104.5	p = 0.01 81.1	93,0	<i>p</i> = 0.00 78.2	906	<i>p</i> =0.00 76.9	94.2	77.1	100.1	77.2	104.7	84.7	96.4	91.4	87.5
		SD	18.9	20.7	12.1	13.7	12.7	12.4	12.7	12.5	14.0	14.9	16.8	16.1	18.6	13.1	19.6	13.3
		N	11	4	12	7	15	13	16	14	18	16	20	19	15	12	8	9
		T-test	t = -2.1;	<i>p</i> = 0.06	t = -2.0; p = 0.06		t = -2.6; p = 0.02		t = -3.8; p = 0.00		t = -4.6;	<i>p</i> =0.00	t = -5.2;	<i>p</i> = 0.00	t = -1.9;	<i>p</i> = 0.08	t=-0.4; p	= 0.69
Expressive	MSEL	Mean	22.2	49.0	40.5	44.7	38.9	45.6	37.9	47.8	40.6	53.4	41.7	53.8	40.7	52.1	48.9	53.5
language		SD	12.5	11.2	6.3	2.8	6.4	6.1	6.1	6.4	10.8	7.3	12.5	7.7	16.4	8.7	11.6	7.2
1		N	11	9	11	10	12	14	16	16	17	17	20	20	15	14	8	9
		T-test	t = -4.4;	p = 0.00	t = -2.0;		t = -2.7;		t = -4.4;		t = -4.0;	p = 0.00	t = -3.7;	p = 0.00	t = -2.4.;	p = 0.03	t = -0.9; p	= 0.41
					<i>p</i> = 0.64		p = 0.01		p = 0.00									
	MCDI	Mean	79.2	115.0	82.7	97.1	88.0	95.9	87.9	97.5	79.9	100.1	75.1	108.2	81.3	126.5	102.8	111.0
		SD	16.6	35.5	15.9	18.3	13.1	11.1	13.7	9.9	12.2	14.5	13.1	20.1	21.0	27.0	30.3	17.9
		N	11	4	12	7	15	13	16	14	18	16	20	19	15	12	8	9
		T-test	t = -1.9;	<i>p</i> =.13	t = -1.8;		t = -1.7;		t = -2.1;		t = -4.4;	p = 0.00	t = -6.1;	p = 0.00	t = -4.9;	p = 0.00	t = -0.6; p	= 0.57
					p = 0.90		p = 0.10		p = 0.39									
N=number of cl:	ildren; SD=	standard dev	/iation: CI = 6	children wit	ch cochlear i	mplant: N	H = children	with norn	hearing:	MSEL = MI	Illen Scale	of Early Lear	ning. Score	are preser	nted as T-sco	ores with a r	ormative mo	ean of 5

l ::

quotients with a normative mean of 100; T-test indicates significance of differences between the Cl group and the normal-hearing group.

as age

presented

Development Inventory. Scores are

MCDI = Minnesota Child

respectively). However, the difference decreased with increasing age and was no longer significant at 36 and 48 months postimplantation, with *T*-scores of 44.1 versus 52.9 and 48.9 versus 49.2, respectively (Table 2). It should be noted that only 14 subjects were tested at the 48-month check-up, and this was also the case for the subsequently mentioned tests performed at the 48-month check-up.

Receptive language scores from the MCDI parent questionnaire Comprehension-Conceptual (CC) subtest, presented in Table 2, show results somewhat similar to those of the MSEL test. At the 3through 24-month check-ups, the CI users achieved scores below one standard deviation from the normative mean. At the 36- and 48-month check-ups, the CI users' mean score approached the normal range. The CI users' mean score at the 3- through 24-month check-up was lower than that of their normal-hearing peers, with the difference reaching statistical significance at the 9- through 24month check-ups (Table 2). The magnitude of the differences in the means was large (eta squared = 0.22, 0.21, 0.22, 0.35, 0.42, at the 3through 24-month check-ups, respectively). However, the difference decreased after 36 months of user time and was not significant at 36 months (84.7 versus 96.4) or at 48 months (91.4 versus 87.5).

# 3.3. Expressive language

Results from the MSEL-expressive language subscale showed that CI users achieved scores close to minus one standard deviation from the normative mean at the 6- through 36-month check-ups (Fig. 2). The CI users' mean scores at the 3- through 48-month check-ups were lower than those of their peers with normal hearing, with the difference reaching statistical significance at 3- and 9 through 36 months of CI use (Table 2). The magnitude of the differences in the means was large (eta squared = 0.59, 0.19, 0.25, 0.41, 0.35, 0.24, at the 3- and 9- through 36-month check-ups, respectively). At 48 months, there was no significant difference between the average scores of the CI users (48.9) and the normally hearing children (53.5) (Table 2).

Results from the MCDI parent questionnaire Expressive Language scale, presented in Table 2, show language scores similar to those found with the MSEL-expressive language tests. The CI users' mean scores at the 3- through 36-month postoperative check-ups varied around minus one standard deviation from the normative mean. The CI users' mean scores at all the data points were lower than their chronological age-matched hearing peers, with the difference reaching statistical significance at the 12-through 36-month check-ups (Table 2). The magnitude of the differences in the means was strong (eta squared = 0.14, 0.39, 0.52, 0.55, at the 12- through 36-month check-ups, respectively). At the 48-month check-up, the CI users' mean score was within the normal range, and there was no significant difference between the mean scores of the CI users (102.8) and the normally hearing children (111) (see Table 2).

# 3.4. Analysis of a subset of subjects

Because not all the children have results at all check-up points, analyses presented in Figs. 1 and 2 includes a different set of subjects at each data point. To minimize subject variability an analysis of a subset of 22 subjects (11 CI users and 11 normalhearing) with complete data from 3 through 24 months postimplantation was done. Fig. 3a and b shows the mean receptive and expressive scores over time on the MSEL for these children. Fig. 3c and d shows CI users' individual receptive and expressive scores. An analysis of language performance showed a significant interaction between the changes over time and group (CI users versus normal hearing), i.e., that the changes over time differed

Receptive and expressive language performance over time in the 21 Cl users and 21 age-matched children with normal hearing.



**Fig. 3.** Average receptive and expressive language performance over time for a subset of 22 subjects (11 Cl users and 11 normal-hearing) who had data points at every time period from 3 through 24 months post-implantation, measured by the Mullen Scales of Early Learning. (a and b) Show the results for normal-hearing children versus Cl users. (c and d) Show individual results for the Cl users.

between the groups: Wilk's lambda = 0.80, F(2,3) = 3.5, p = 0.04, multivariate eta squared = 0.20. Repeated-measure ANOVA was further used to examine changes over time on the MSEL receptive and expressive language subscale scores of the CI users at 3, 6, 9, 12, 18, and 24 months post-implantation. For both receptive and expressive language, the results showed a significant time effect: Wilk's lambda = .22, F = 4.4, p = 0.05 and Wilk's lambda = 0.16, F = 6.2, p = 0.02, respectively. The mean *T*-scores improved significantly between 3 and 24 months, with increases from 28 to 47 for receptive language and from 22 to 44 for expressive language. Thus, over a user time of 24 months, the mean score approached the range of  $\pm 1$  standard deviation from the normative mean. Note that the individual scores presented in Fig. 3c and d show a large variation within the CI group, with receptive language T-scores ranging from 34 to 64 and expressive language T-scores ranging from 24 to 70.

# 3.5. The relationship between age at connection and language development

For both receptive and expressive language, the results showed significant strong and moderate positive associations with early age at connection and better language scores. However, the associations were strongest for expressive language at the early check-ups, diminished with increased user time, and were no longer significant after 36 months of user time. For receptive language, a moderate-to-strong significant correlation existed between age at connection and MSEL scores at check-up after 3 months (-0.80, p = 0.00) and for the MCDI-CC, at 3 months (-0.61, p = 0.01), and 12 months (-0.65, p = 0.00) user time. For expressive

language, the correlations with age at connection were significant for the MSEL at 6 months (-0.85, p = 0.00), 9 months (-0.68, p = 0.01), 12 months (-0.64, p = 0.00), 18 months (-0.51, p = 0.36), and 24 months (-0.47, p = 0.04) and for the MCDI at 3 months (-0.66, p = 0.02), 9 months (-0.59, p = 0.19), 12 months (-0.65, p = 0.00), and 18 months (-0.66, p = 0.00).

When comparing the language performance and age of implantation, the children who were implanted before 12 months of age had the highest average scores at all times of testing. A linear mixed model for repeated measurements with fixed terms for study groups (children implanted before or after 12 months of age) and observation time showed that children implanted before 12 months of age (N = 13) had significantly higher scores than those implanted between 12 and 18 months of age (N = 8). Mean MSEL receptive language scale scores were 44.6 versus 34.6: (95% CI: 0.80-19.17, p = 0.035). MSEL-expressive language scale means were 43.0 versus 32.3: 10.7 (95% CI: 3.4–18.0, p = 0.007). Fig. 4 shows that the mean scores of the children implanted before age 12 months stabilized within  $\pm 1$  SD from the normative mean after 12 months of use for receptive language and after 18 months of use for expressive language. In contrast, the mean scores of the children implanted between 12 and 18 months of age first reached the range of normal development for both receptive and expressive language after 48 months of CI use.

# 4. Discussion

#### 4.1. Language issues

The present study is one of the first to offer long-term expressive and receptive spoken language outcome data from a



Fig. 4. Average receptive and expressive language performance over time in children who received CI before 12 months of age versus children who received CI between 12 and 18 months of age, measured by the Mullen Scales of Early Learning. It should be noted that not all the subjects have data at each data point.

group of prelingually deaf children who received simultaneous bilateral cochlear implantation between the ages of 5 and 18 months. As of yet, there are few children who have received this treatment. This study indicates that 81% of these children developed receptive language skills and 57% developed expressive language skills inside the normative range within 12–48 months after implantation.

Functional language skills need several years to develop and can be challenging for prelingually deaf children with cochlear implants. The risk of delays and limitations in spoken language development are apparent; spoken language skills might develop according to the child's hearing age, and/or at a slower rate than expected according to how long the child has had access to sound. This indicates delayed language development and/or a gap between normal-hearing children's and CI users' language skills that increases with age. This was true for 19% of the children in the present study, who had receptive and expressive scores at or below the second percentile. All of these children were in mainstream education. This finding indicates that children with early and bilateral implantation need long-term language monitoring aimed at identifying each child's individual educational and social needs.

Another language development scenario for the prelingually deaf children is that after receiving CIs, the child will develop language skills at a faster pace than their hearing age would indicate and thereby attain age-equivalent language skills. Evidence of increased rate of development compared to normalhearing children has been found in studies of children with early unilateral cochlear implantation [2,22,23]. The current paper reveals that this scenario is true for most of the early implanted, bilateral CI users in this study. Receptive and expressive language test scores demonstrated that the majority of the children with implants in this study had a faster pace of language development than their hearing age would indicate. Over time, they attained age-equivalent language skills and decreased the language gap between them and their normal-hearing pears.

In earlier research performed on unilateral and laterimplanted children (>12 months) that scored within the normative range there was evidence for significantly lower scores among the CI users compared to their normal-hearing pairs [24,27]. Although this was true at some of the data points in the present study, our findings diverge somewhat from these findings as the difference in scores decreased with increased user time and were no longer significant after 36 months with CI for receptive language (p = 0.08) and after 48 months for expressive language (p = 0.41). The above mentioned studies [24,27] were cross-sectional studies and it is possible that some of the lower results can be accounted for by the inclusion of older and later-implanted children and by unilateral (the lack of auditory binaural perception advantages) versus bilateral implantation.

In comparison with earlier studies, we observed an increased number of children attaining language skills within the normal range at a younger age. According to Hart and Risley [4], hearing experience in early childhood lays the foundation for more advanced use and acquisition of language. It is possible that the age at which a child reaches the normal range of language performance may be important to further language development. After 12 months of CI experience and an average chronological age of 25 months, 38% of the children with CI had receptive scores within the normal range, and 44% had expressive language scores within the normal range. In the normal-hearing group 94% scored within the normative range on both receptive and expressive language. After 24 months of CI use and an average chronological age of 36 months, 60% of children with CI had receptive and 55% had expressive scores within the normal range, whereas 90 and 100% of the normal-hearing children had scores within the normative range in receptive and expressive language respectively. In comparison, Geers et al. [25] studied 5and 6-year-old prelingually deaf children, mostly unilateral implanted, and found that 47% attained age-appropriate receptive language scores and 39% attained age-appropriate expressive language scores. In that study, children who received their implants at young ages had higher scores on all language tests than those implanted at older ages.

Our result of a higher increase in the proportion of children with receptive as opposed to expressive scores within the normative range shows that the children with CI need more time to develop age equivalent expressive language. The finding compares to earlier studies findings that unilateral children's development of expressive language skills to be much slower and/or variable than their development of receptive language as opposed to receptive language were not suitable for predicting later language performance. With 81% of the children in the present study falling within the normal range of receptive language skills during a CI user time of 12–48 months, our findings seem promising indicators for the children's later language performance.

# 4.2. Early identification and eventual secondary issues

One issue with early identification of hearing loss is that at the time of enrollment in a particular study, a child may initially appear to have a normal developmental pattern, yet additional disabilities may surface as time elapses. This appeared to be the case for three children in this study, all with results at or below the second percentile. In these cases, further investigation has been implemented due to suspicion of implant complications or additional disabilities in the child.

The children's language was considered to be developing if they achieved approximately equal *T*-scores after 18, 24 and 36 months. This rate of development may indicate that a child's rate of language development will remain below that of their hearing peers for reasons which we do not yet have plausible explanations. Some of the reasons might not be directly related to having a hearing loss. Such secondary issues could involve different kinds of medical diseases and conditions, or also language disorders like specific language impairment or an auditory processing disorder. Normal-hearing children with specific language impairment will have unexpected and unexplained difficulties learning and using spoken language [26]. Specific language impairment could also occur in the population of CI users. It is likely that they will experience similar difficulties as normal-hearing children with specific language impairment.

These types of scenarios may be common when children receive implants before 12 months of age, as presumably there will be some who have additional difficulties that appear as they grow older. It is crucial that the instructions given to the parents and educators explain that further developmental issues may occur. Additionally, to ensure adequate follow-up over time, it is essential to study a wide spectrum of language skills, such as vocabulary use in coherent speech, understanding of syntax, and grammatical constructions.

# 4.3. The contribution of variation and implications for future studies

Our results are in agreement with a number of other studies that show a significant correlation between age at implantation and scores both in hearing development as well as expressive and receptive language in unilateral implanted children [2,10,11,23,28,29]. It is noteworthy that even though all children in this study received bilateral CIs between 5 and 18 months of age, age at implantation still had an effect on language development. In line with studies of unilateral implantation in children under 12 months old [2,10,11,30,31,32], the children in this study who were implanted between 5 and 12 months attained higher scores and had a significant lead in reaching the normal language range than children who received implants between 12 and 18 months of age. It seems, however, that the effect of early implantation is greatest in the first postoperative years. It is reasonable to assume that a number of other factors, such as the characteristics of the child and factors connected with habilitation, will be of significance and will emerge as the child grows older [7,33,34]. In this study, there was no direct control over the degree of speech and language stimulation, the mode of communication, or the form of interaction in the child's surroundings. Thus, variations in these factors may be important contributors to differences in development. All the parents had received shorter or longer instructions based on auditory oral or auditory verbal training, but again it is likely that variations in interactional patterns had occurred. Based on the findings of Hart and Risley [4], it is likely that different interactional patterns manifest themselves among CI users as among normalhearing children and contributes to variation in language skills. Thus, a possible hypothesis is that implantation within a certain age (in this study within 18 months) is a prerequisite for age equivalent speech and language development but that the effect of age at implantation will become less evident as other factors become more important over time.

#### 4.4. Study limitations

The number of participants in this study was limited, as the population of children with simultaneously bilateral cochlear implantation between the 5th and 18th month is still small. The data are somewhat limited, as complete test results could not be obtained at all data points for all of the children. The results at the 48-month check-up must be considered an early indication and should be interpreted cautiously. Further follow-ups until these children have reached 72 months of experience are planned, and the test battery will be supplemented with more extensive tests, including a test of understanding grammatical construction in language.

#### 5. Conclusions

The present study showed that prelingually deaf children's ability to develop complex expressive and receptive spoken language after early, bilateral implantation appears promising. The majority of the children in this study developed language skills at a faster pace than their hearing age would suggest. The results indicated that the majority of the children had expressive and receptive language skills within the normative range after 12–48 months of Cl use. The proportion of children within the normative range increased with implant experience.

# **Conflict of interest**

The author has no conflicts of interest.

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### References

- M.P. Moeller, Current state of knowledge: language and literacy of children with hearing impairment, Ear Hear. 28 (6) (2007) 740–753, Review.
- [2] S.J. Dettman, D. Pinder, R.J. Briggs, R.C. Dowell, J.R. Leigh, Communication development in children who receive the cochlear implant younger than 12 months: risks versus benefits, Ear Hear. 28 (2007) 11–18.
- [3] K.I. Kirk, R.T. Miyamoto, C.L. Lento, E. Ying, T. O'Neill, B. Fears, Effects of age at implantation in young children, Ann. Otol. Rhinol. Laryngol. Suppl. 189 (2002) 69– 73.
- [4] B. Hart, T.R. Risley, The Social World of Children Learning to Talk, Brookes Publishing Co., Baltimore, 1999.
- [5] C.E. Snow, O.P. Tabors, D.K. Dickinson, Language development in the preschool years, in: D.K. Dickinsom, P.O. Tabors (Eds.), Beginning Literacy with Language, Baltimore Brookes Publishing Co., 2001, pp. 1–25.
- [6] LJ. Spencer, J.J. Oleson, Early listening and speaking skills predict later reading proficiency in pediatric cochlear implant users, Ear Hear. 29 (2) (2008) 270–280.
- [7] M.J. Hay-McCutcheon, K.I. Kirk, S.C. Henning, S. Gao, R. Qi, Using early language outcomes to predict later language ability in children with cochlear implants, Audiol. Neurootol. 13 (6) (2008) 370–378.
- [8] C.M. Connor, H.K. Craig, S.W. Raudenbush, K. Heavner, T.A. Zwolan, The age at which young deaf children receive cochlear implants and their vocabulary and speech-production growth: is there an added value for early implantation? Ear Hear. 27 (6) (2006) 628–644.
- [9] M. Tait, L. De Raeve, T.P. Nikolopoulos, Deaf children with cochlear implants before the age of 1 year: comparison of preverbal communication with normally hearing children, Int. J. Pediatr. Otorhinolaryngol. 71 (2007) 1605–1611.
- [10] T.Y. Ching, H. Dillon, J. Day, Early language outcomes of children with cochlear implants: interim findings on the NAL study on longitudinal outcomes of children with hearing impairment, Cochlear Implants Int. 10 (2008) 28–32.
- [11] R.T. Miyamoto, M.J. Hay-McCutcheon, K.I. Kirk, D.M. Houston, T. Bergerson-Dana, Language skills of profoundly deaf children who received cochlear implants under 12 months of age: a preliminary study, Acta Otolaryngol. 128 (4) (2008) 373–377.

- [12] M.N. Ruscetta, E.M. Arjmand, S.R. Pratt, Speech recognition abilities in noise for children with severe-to-profound unilateral hearing impairment, Int. J. Pediatr. Otorhinolaryngol. 69 (6) (2005) 771–779.
- [13] K.D. Brown, T.J. Balkany, Benefits of bilateral cochlear implantation: a review, Curr. Opin. Otolaryngol. Head Neck Surg. 5 (5) (2007) 315–318.
- [14] K.A. Gordon, B.C. Papsin, Benefits of short interimplant delays in children receiving bilateral cochlear implants, Otol. Neurotol. 30 (3) (2009) 319–331.
- [15] J.C. Johnston, A. Durieux-Smith, D. Angus, A. O'connor, E. Fitzpatrick, Bilateral paediatric cochlear implants: a critical review, Int. J. Audiol. 48 (9) (2009) 601–617.
- [16] S.A. Chang, R.S. Tyler, C.C. Dunn, H. Ji, S.A. Witt, B. Gantz, M. Hansen, Performance over time on adults with simultaneous bilateral cochlear implants, J. Am. Acad. Audiol. 21 (1) (2010) 35–43.
- [17] M. Tait, T.P. Nikolopoulos, L. De Raeve, S. Johnson, G. Datta, E. Karltorp, et al., Bilateral versus unilateral cochlear implantation in young children, Int. J. Pediatr. Otorhinolaryngol. 74 (2) (2010) 206–211.
- [18] H. Kühn-Inacker, V. Weichbold, L. Tsiakpini, F. Coninx, P. D'Haese, LittlEARS Auditory Questionnaire Manual–Parent Questionnaire to Assess Auditory Behavior in Young Children, MED-EL, Innsbruck, 2003.
- [19] E. Mullen, Manual for the Mullen Scales of Early Learning: AGS Edition. Circle Pines, American Guidance Service Inc., MN, 1995.
- [20] H. Ireton, E. Thwing, Manual for the Minnesota Child Development Inventory, Behavior Science Systems, Minneapolis, 1974.
- [21] J.B. Tomblin, C.M. Shonrock, J.C. Hardy, The concurrent validity of the Minnesota Child Development Inventory as a measure of young children's language development, J. Speech Hear. Disord. 54 (1) (1989) 101–105.
- [22] H. Hayes, A.E. Geers, R. Treiman, Receptive vocabulary development in deaf children with cochlear implants: achievement in an intensive auditory-oral educational setting, Ear Hear. 30 (1) (2009) 128–135.
- [23] J.K. Niparko, E.A. Tobey, D.J. Thal, L.S. Eisenberg, N.Y. Wang, A.L. Quittner, N.E. Fink, CDaCl Investigative Team, Spoken language development in children following cochlear implantation, JAMA 21 (303 (15)) (2010) 1498–1506.

- [24] E.A. Schoor, F.P. Roth, N.A. Fox, A comparison of the speech and language skills of children with cochlear implants and children with normal hearing, Comm. Disord. Q. 29 (2008) 195.
- [25] A.E. Geers, J.S. Moog, J. Biedenstein, C. Brenner, H. Hayes, Spoken language scores of children using cochlear implants compared to hearing age-mates at school entry, J. Deaf. Stud. Deaf. Educ. 14 (3) (2009) 371–385.
- [26] J.B. Tomblin, The EpiSLI database: a publicly available database on speech and language, Lang. Speech Hear. Serv. Sch. 41 (1) (2010) 108–117.
- [27] J.R. Inscoe, A. Odell, S. Arcbold, T. Nikolopoulos, Expressive spoken language development in deaf children with cochlear implants who are beginning formal education, Deaf. Educ. Int. 11 (2009) 39–55.
- [28] D.M. Hammes, M.A. Novak, L.A. Rotz, M. Willis, D.M. Edmondson, J.F. Thomas, Early identification and cochlear implantation: critical factors for spoken language development, Ann. Otol. Rhinol. Laryngol. Suppl. 89 (2002) 74–78.
- [29] B.J. Tomblin, B.A. Barker, S. Hubbs, Developmental constraints on language development in children with cochlear implants, Int. J. Audiol. 46 (2007) 512– 523.
- [30] L. Colletti, Long-term follow-up of infants (4–11 months) fitted with cochlear implants', Acta Oto-Laryngol. 129 (4) (2009) 361–366.
- [31] P.V. Vlastarakos, K. Proikas, G. Papacharalampous, I. Exadaktylou, G. Mochloulis, T.P. Nikolopoulos, Cochlear implantation under the first year of age—the outcomes. A critical systematic review and meta-analysis, Int. J. Pediatr. Otorhinolaryngol. 74 (2) (2010) 119–126.
- [32] M. Cosetti, J.T. Roland Jr., Cochlear implantation in the very young child: issues unique to the under-1 population, Trends Amplif. 14 (1) (2010) 46–57.
- [33] A. Geers, C. Brenner, L. Davidson, Factors associated with development of speech perception skills in children implanted by age five, Ear Hear. 24 (1) (2003) 24–35.
- [34] O.B. Wie, E.S. Falkensberg, O. Tvete, B.J. Tomblin, Children with cochlear implant: characteristics and determinants of speech recognition, speech-recognition growth rate, and speech production, Int. J. Audiol. 46 (2007) 232–243.