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Long-term effects of second cochlear implantation with sequential bilateral cochlear implantation in Japanese children

Naoe Mori[a,b], Norio Yamamoto[a,c,*], Shinobu Yamaguchi[a,d], Kanako Kondo[a], Misaki Yoshizawa[a], Takayuki Okano[a], Juichi Ito[e], Koichi Omori[a]

[a] *Department of Otolaryngology Head and Neck Surgery, Graduate School of Medicine, Kyoto University, 54 Shogoin Kawahara-cho, Sakyo-ku, Kyoto City, Kyoto 606-8507, Japan*
[b] *Department of Rehabilitation Sciences, Faculty of Allied Health Sciences, Kansai University of Welfare Sciences, Osaka, Japan*
[c] *Department of Otolaryngology, Kobe City Medical Center General Hospital, Japan*
[d] *Osaka Health Science University, Osaka, Japan*
[e] *Shiga General Hospital, Shiga, Japan*

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ABSTRACT

Objective: This study aimed to longitudinally evaluate speech perception ability and sound-field thresholds with the first, second, or bilateral cochlear implants (CIs) and MAP parameters of second CI in children.
Methods: Eighteen children who underwent bilateral cochlear implantation at Kyoto University Hospital were included. We evaluated speech perception under quiet and noisy conditions using the first, second, or bilateral CIs, CI-aided sound-field thresholds using the first or second CI, and MAP parameter values (C-levels, T-levels, and dynamic range) of the second CI of more than 5 years after the second implantation.
Results: Patients with a second CI after 7 years of age had significantly worse speech perception ability with the second CI even long after the surgery than those with a second CI before 7 years of age. CI-aided sound-field thresholds using the first or second CI were similar, regardless of the second implantation timing. Speech perception in noise with bilateral CIs was enhanced by the addition of a second CI, even after 7 years of age. Patients undergoing second cochlear implantation before 3.5 years of age showed significantly higher C-levels and wider dynamic ranges in the second CI MAP parameters.
Conclusions: When the second implantation was performed after 7 years of age, the second CI effects were limited even with long-term use, which is attributed to unstable MAP parameters. The second CI-aided sound-field threshold contributed to the better outcome of bilateral CIs in noise, even if the second implantation was performed at age of ≥7 years.

1. Introduction

The effectiveness of bilateral cochlear implantation in improving hearing in noise and sound localization has been clarified [1–5]. However, when bilateral cochlear implants (CIs) are implanted sequentially, the effects of the second CI and the benefits of bilateral hearing vary

* Corresponding author at: Department of Otolaryngology Head and Neck Surgery, Graduate School of Medicine, Kyoto University, 54 Shogoin Kawahara-cho, Sakyo-ku, Kyoto City, Kyoto 606-8507, Japan.
E-mail address: yamamoto@ent.kuhp.kyoto-u.ac.jp (N. Yamamoto).

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depending on the inter-implant interval and age at second implantation [6–9].

After the revised Japanese selection criteria for pediatric cochlear implantation supported bilateral cochlear implantation in 2014, children with unilateral CI could undergo sequential bilateral cochlear implantation in Japan.

We previously investigated the relationship between the effects of sequential bilateral cochlear implantation and age at the second implantation by analyzing speech perception ability and sound localization. The second CI was found to be beneficial if implanted at 7 years of age or younger. However, no significant difference was observed in speech perception in noise between unilateral and bilateral CIs 1–5 years (2 years 6 months on average) after the second implantation [10]. Considering the possibility of the slow development of bilateral effects, we decided to collect data at much later stages and at several time points to reveal the long-term outcomes after the second implantation.

The present study examined longitudinal changes in speech perception ability and sound-field thresholds in children with long-term post-implantation follow-up after sequential bilateral cochlear implantation. Additionally, we examined MAP parameter values, including comfort levels (C-levels), threshold levels (T-levels), and dynamic range, of the second CI to investigate the relationship between CI performance and the long-term outcomes of cochlear implantation.

2. Materials and methods

2.1. Patient characteristics

This study included 18 children (nine men and nine women) aged 5 years 6 months to 16 years 2 months (mean, 10 years 11 months; median, 10 years 1 month). All of them used their second CI for more than 3 years (mean, 6 years 6 months; median, 7 years). The children underwent sequential bilateral cochlear implantation using Cochlear Ltd. devices at Kyoto University Hospital from 2005 to 2017. The cochlear nerve hypoplasia cases were excluded based on pre-operative MRI images. Patients had prelingual deafness and did not have delays in language development, as demonstrated by scores of 80 or higher on various tests for language development. The tests used in this study included the development quotient in the language-social domain of the 2001 Kyoto Scale of Psychological Development and the verbal comprehension index calculated based on the Wechsler Preschool and Primary Scale of Intelligence-III and the Wechsler Intelligence Scale for Children-IV.

As in our previous study [10], patients were classified into three groups based on age at second implantation: patients in group A underwent second cochlear implantation before 3.5 years of age (n = 9). Group B (n = 6) and C (n = 3) included patients who underwent second cochlear implantations between 3.5 and 7 years of age and at more than 7 years of age, respectively. There were several different reasons for the delayed second cochlear implantation. Two group B and all three group C patients underwent their first cochlear implantation more than several years before 2014 when the revised Japanese selection criteria for pediatric cochlear implantation supported bilateral cochlear implantation. Two group B patients underwent the second implantation before enrolling in the elementary school with a noisier environment than their home. The other two group B patients decided to undergo the second implantation after several years of wavering. The mean age at the time of first cochlear implantation in groups A, B, and C was 1 year 9 months, 1 year 9 months, and 2 years 2 months, respectively. Meanwhile, the mean ages of patients undergoing the second implantation in groups A, B, and C were 2 years 7 months, 5 years 2 months, and 8 years 5 months, respectively.

The causes of deafness were unknown in all but two patients with GJB2 mutations on genetic testing. All patients wore hearing aids before the first and second cochlear implantations. The implanted devices were CI24M (n = 1), CI24R (n = 4), CI24RE (CA) (n = 12), or CI512 (n = 1) for the first CI (first CI) and CI24RE (CA) (n = 10), CI422 (n = 4), CI512 (n = 2), or CI522 (n = 2) for the second CI. The sound processors used by the patients were bilateral CP810 (n = 2), CP810 and CP910 (n = 5), CP810 and CP1000 (n = 1), bilateral CP910 (n = 6), CP910 and CP1000 (n = 3), and bilateral CP1000 (n = 1). Default MAP parameter settings were used for all the patients. Four cases had unused one or two electrodes among 22 electrodes. The number of unused electrodes was as follows: two in bilateral CIs, n = 1 (group C); two in the first CI and one in the second CI, n = 1 (group A); two in the first CI, n = 1 (group B); and one in the first CI, n = 1 (group B) (Table 1).

2.2. Sound-field thresholds and speech perception tests in quiet and noise

CI-aided sound-field threshold tests were performed using the peep show test or sound-field test. The mean sound-field thresholds were calculated by averaging the thresholds at 0.5, 1, 2, and 4 kHz. Speech perception tests were performed under quiet and noisy conditions. Two loudspeakers were arranged in front, and each loudspeaker was set at the level of the child's left and right external auditory canals and used for speech perception tests. Speech and noise were presented from two loudspeakers. Japanese monosyllabic words (67-S list, one list: 20 sounds) were presented at 60 dB HL from a distance of 1 m. The children were asked to respond by repeating the given words with speech or dictation, and the rate of correct answers was calculated from the number of correct responses. All tests were conducted in an anechoic room. The noise was presented as previously described [10], with a speech signal at a signal-to-noise ratio of +10 dB. The audiometer used in this study was calibrated according to ISO389-2 to show dB SPL-based data in dB HL. CI-aided sound-field threshold tests were performed unilaterally with the first CI alone or the second CI alone under quiet conditions, whereas speech perception tests were performed under quiet and noisy conditions wearing bilateral CIs, the first CI alone, or the second CI alone.

These tests were performed every year for 6 years after the second implantation (years 1–6: 1y–6y) and we compared

Table 1. Patient characteristics.

Group	Mean age (Median; Range)	Mean age at 1st CI implantation (Median; Range)	Mean age at 2nd CI implantation (Median; Range)	Device	The numbers of unused electrodes	Processor
Group A (9 patients)	8 y 8 m (8 y 7 m; 5 y 6 m – 11 y 2 m)	1 y 9 m (1 y 6 m; 1 y 3 m – 2 y 6 m)	2 y 7 m (2 y 6 m; 1 y 9 m – 3 y 2 m)	Bilateral CI24RE(CA) (n = 5) CI24RE(CA) and CI422 (n = 2) CI24RE(CA) and CI512 (n =1) CI512 and CI522 (n = 1)	Two electrodes in 1st CI and one electrode in 2nd CI (1 patient)	Bilateral CP810 (n = 1) CP810 and CP910 (n = 2) CP810 and CP1000 (n = 1) Bilateral CP910 (n = 3) CP910 and CP1000 (n = 2)
Group B (6 patients)	11 y 10 m (13 y 1 m; 8 y 2 m – 14 y 9 m)	1 y 9 m (1 y 9 m; 1 y 6 m – 2 y 4 m)	5 y 2 m (6 y 2 m; 3 y 8 m – 6 y 10 m)	CI24R and CI24RE(CA) (n = 2) Bilateral CI24RE(CA) (n = 2) CI24RE(CA) and CI512 (n = 1) CI24RE(CA) and CI522 (n = 1)	One electrode in 1st CI (1 patient) Two electrodes in 1st CI (1 patient)	Bilateral CP810 (n = 1) CP810 and CP910 (n = 1) Bilateral CP910 (n = 3) CP910 and CP1000 (n = 1)
Group C (3 patients)	15 y 9 m (15 y 11 m; 15 y 1 m – 16 y 2 m)	2 y 2 m (2 y 6 m; 1 y 2 m – 2 y 10 m)	8 y 5 m (8 y 7 m; 7 y 10 m – 8 y 10 m)	CI24R and CI24RE(CA) (n = 1) CI24R and CI422 (n = 1) CI24M and CI422 (n = 1)	Two electrodes in 1st CI and two electrodes in 2nd CI (1 patient)	CP810 and CP910 (n = 2) Bilateral CP1000 *n = 1)

m, months; y, years. All devices and processors are those from Cochlear Ltd.

inter- and intragroup outcomes when patients wore the first CI alone, the second CI alone, or bilateral CIs. However, the number of patients was not constant at each time point. Under the quiet condition, the patient numbers are as follows: Group A (1y: n = 4, 2y: n = 9, 3y: n = 9, 4y: n = 7, 5y: n = 6, 6y: n = 6), group B (1y: n = 5, 2y: n = 6, 3y: n = 6, 4y: n = 6, 5y: n = 4, 6y: n = 4), and group C (1y–6y: n = 3). Under the noise condition, the patient numbers are as follows: Group A (1y: n = 4, 2y: n = 4, 3y: n = 8, 4y: n = 6, 5y: n = 6, 6y: n = 6), group B (1y: n = 4, 2y: n = 6, 3y: n = 6, 4y: n = 6, 5y: n = 4, 6y: n = 4), and group C (1y–6y: n = 3).

The mean and median duration between the second implantation and the latest examination was 6 years 1 month and 6 years 4 months (3 years–8 years 5 months) for group A, 6 years 8 months and 6 years 7 months (4 years–10 years) for group B, and 7 years 4 months and 7 years 3 months (7 years 1 month to 7 years 7 months) for group C.

2.3. MAP parameters of the second CI

To investigate CI performance, MAP parameter values (C-levels, T-levels, and dynamic range) of the second CI at 1 and 5 years after the second implantation were collected from medical records (n = 14. Group A, n = 7; group B, n = 4; and group C, n = 3). The amount of change between these two timings was calculated to elucidate the long-term MAP changes. The C-levels, T-levels, and dynamic ranges were examined for all electrodes used in the patients.

2.4. Statistics

Differences in each parameter were examined using Kruskal-Wallis one-way analysis of variance with post-hoc Steel-Dwass or Mann-Whitney U test. The aligned rank transform [11,12] with the post-hoc Steel-Dwass test was used to conduct a nonparametric two-way analysis of variance. A p-value less than 0.05 was considered as significant. All statistical analyses were conducted using SPSS version 24.0 (IBM Corp, Armonk, NY, USA) or R version 4.1.2 (https://www.r-project.org/), using the ARTool package (https://cran.r-project.org/web/packages/ARTool/index.html).

2.5. Ethics approval

This study was approved by the Kyoto University Graduate School and Faculty of Medicine Ethics Committee (R0842) and was performed in accordance with the Helsinki Declaration.

3. Results

3.1. Speech perception in quiet and in noise

Under the quiet condition, group C showed a significantly worse speech perception than group A when wearing the second CI alone at the latest examination (Fig. 1a, Table 2, p = 0.007). The speech perception ability of patients in groups A and B was comparable when they wore bilateral CIs or the first CI alone. When group A used the second CI alone, the results of the speech perception test were stable

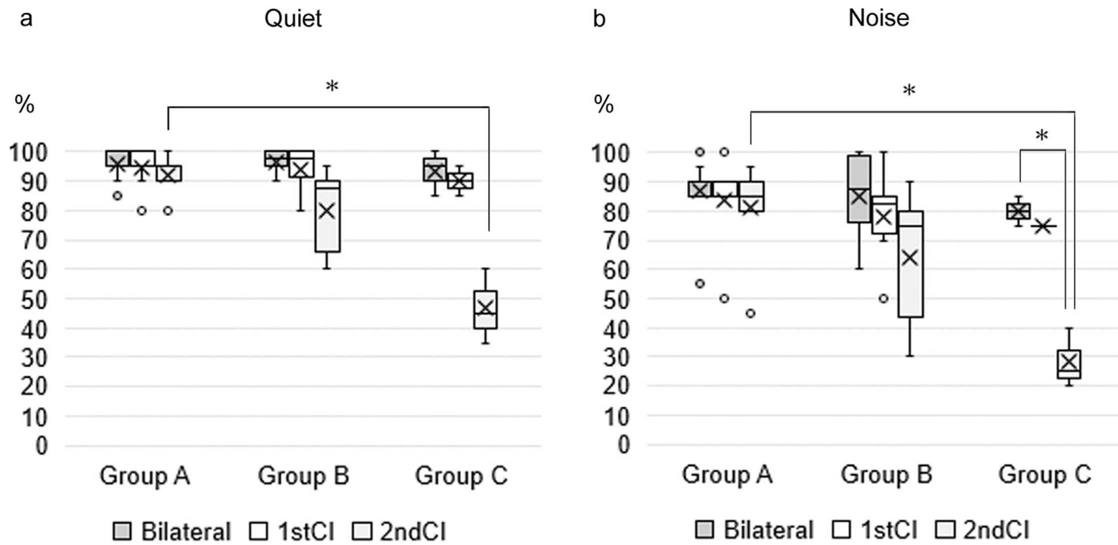

Fig. 1. Speech perception in bilateral cochlear implant users under quiet and noise conditions. The mean correct rate (%) for Japanese speech perception in bilateral cochlear implant users (a: in quiet and b: in noise) is evaluated using Japanese monosyllabic words. The limits of the box represent the lower and upper quartiles of the distribution (the difference is the interquartile range [IQR]). The horizontal line through the box and X represents the median and mean, respectively. The whiskers represent the minimum and maximum scores in the distribution, excluding outliers. Open-circle outliers are values between 1.5 and 3 IQRs from the end of a box. Speech perception is significantly lower in group C than that in group A when wearing the second CI under quiet and noise conditions. Within group C, speech perception is significantly lower in noise with the second CI alone than with bilateral CIs. * $p < 0.05$.

Table 2. Speech perception (Japanese monosyllabic words).

		Bilateral (Mean±SD)	1st CI (Mean±SD)	2nd CI (Mean±SD)
Group A	Quiet	95.6±5.0	94.4±6.0	92.2±5.8
	Noise	86.7±12.0	83.9±12.9	81.1±13.7
Group B	Quiet	96.7±3.7	94.2±7.3	80.0±14.4
	Noise	85.0±14.7	78.3±15.5	64.2±23.2
Group C	Quiet	93.3±6.2	90.0±4.1	46.7±10.3
	Noise	80.0±4.1	75.0±0.0	30.0±8.5

Mean correct rate±standard deviation (SD) (%) for Japanese monosyllabic speech perception on bilateral, first, and second CIs under each group and condition (quiet or noise) at the latest examination. Noise; signal-to-noise ratio of +10 dB.

and comparable with those with bilateral CIs or the first CI alone. However, patients in group B with the second CI alone showed variable speech perception test results. In group C, the second CI usage showed worse results for speech perception than the bilateral or first CI usage, although the difference was not statistically significant.

Under the noisy condition, the rate of correct answers with the second CI alone was significantly worse in group C than in group A (Fig. 1b, Table 2, $p = 0.004$), as observed under the quiet condition. The rate of correct answers with bilateral CIs was better than that with the first CI alone or second CI alone. However, the difference was not significant except the comparison between the bilateral CIs and second CI alone in group C (Fig. 1b, Table 2, $p = 0.018$).

Next, we examined the differences in speech perception ability with the second CI alone among the three groups longitudinally (Fig. 2). Significant differences in the rate of correct answers with the second CI alone were observed between groups A and C ($p < 0.0001$) and between groups B and C ($p < 0.0001$) under the quiet condition (Fig. 2a). Under the noisy condition, significant differences were observed between groups A and B ($p = 0.0017$), groups A and C ($p < 0.0001$), and groups B and C ($p < 0.0001$). The interaction between CI wearing side and the time course was not significant in this analysis.

In group A, no significant differences in the rate of correct answers were observed among using bilateral CIs, the first CI alone, and the second CI alone under quiet and noisy conditions (Fig. 3). In group B, wearing bilateral CIs showed significantly better speech perception scores than wearing second CI alone in quiet ($p = 0.0062$), first CI alone in noise ($p = 0.0118$), and second CI alone in noise ($p < 0.0001$) (Fig. 3). In group C, wearing the second CI alone showed significantly worse speech perception scores compared with wearing bilateral CIs ($p < 0.0001$) and the first CI alone ($p < 0.0001$) under the quiet condition. For noise, significant differences were observed between bilateral CIs and second CI alone ($p < 0.0001$), first CI alone and second CI alone ($p < 0.0001$), and bilateral CIs and first CI alone ($p = 0.0012$) (Fig. 3). The interaction was not significant in this analysis.

3.2. CI-aided sound-field thresholds

While CI-aided sound-field thresholds (Fig. 4) were slightly better with the first CI alone in group C than in groups A and B, this intergroup difference was marginal at

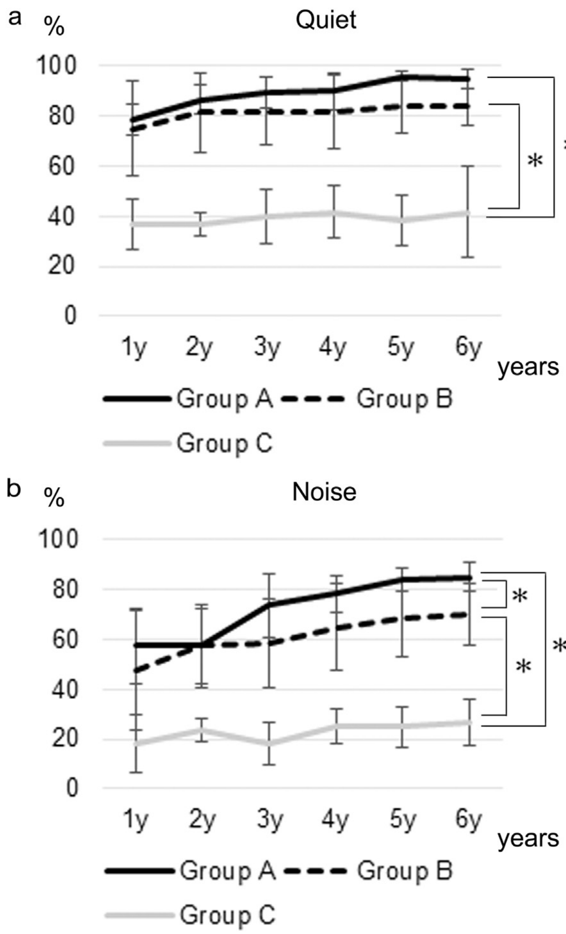

Fig. 2. Longitudinal change in the speech perception wearing the second CI alone in quiet and noise. The mean rate of correct answers ± standard deviation (SD) (%) for speech perception wearing the second CI alone is plotted every year for 6 years after the second implantation using Japanese monosyllabic words in quiet (a) and noise (b). Speech perception in group C is significantly lower than that in groups A and B under quiet conditions. Speech perception of noise shows a significant difference in all the groups. * $p < 0.05$.

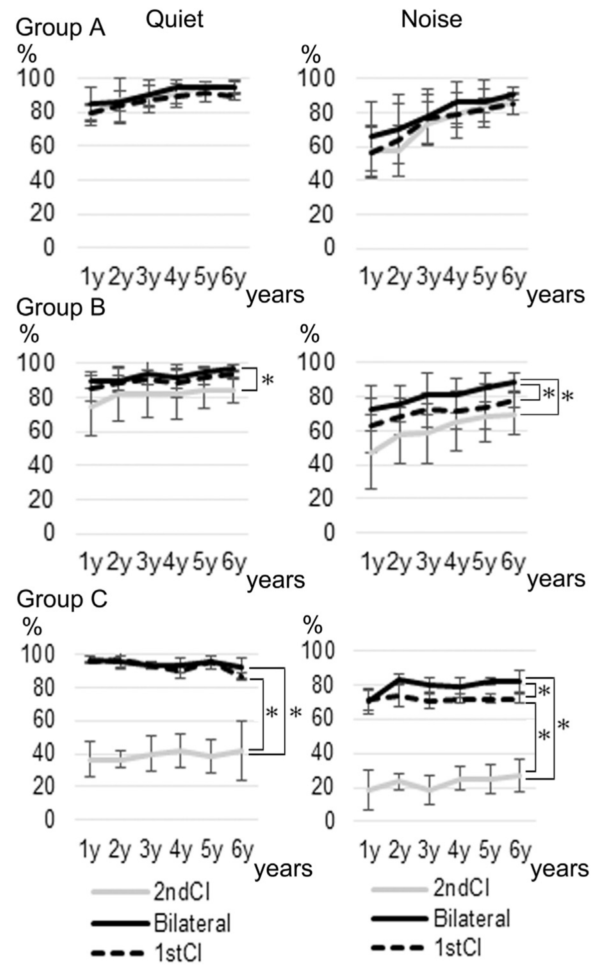

Fig. 3. Speech perception in quiet and noise. The mean rate of correct answers ± standard deviation (SD) (%) for speech perception with bilateral CIs, the first CI alone, or the second CI alone under quiet and noise conditions is longitudinally presented for groups A, B, and C patients. Group A patients have similar speech perception in quiet and noise conditions, regardless of the type of CI usage. In contrast, using bilateral CIs in group B patients resulted in better speech perception than using the second CI alone under quiet conditions and using the first CI or second CI under noisy conditions. Group C patients have lower speech perception under both quiet and noisy conditions when wearing the second CI alone compared with other CI usage. Additionally, even the speech perception while wearing the first CI alone is lower than that while wearing bilateral CIs in noise in group C. * $p < 0.05$.

Table 3. Cochlear implant aided sound-field thresholds of bilateral CIs patients.

	1st CI (Mean±SD)	2nd CI (Mean±SD)
Group A	30.1±4.1	29.9±4.7
Group B	27.9±4.5	30.0±7.4
Group C	25.6±2.1	28.1±3.9

The mean thresholds±SD (dBHL) on first and second CI in each group at the latest examination.

the latest examination (Fig. 4a, Table 3). In group A, the CI-aided sound-field thresholds with the first CI alone and the second CI alone were almost identical (Fig. 4a, Table 3). In group B, a better CI-aided sound-field threshold was observed with the first CI alone, although the difference was not statistically significant (Fig. 4a, Table 3). In group C, a slightly better CI-aided sound-field threshold was obtained with the first CI alone than with the second CI alone (Fig. 4a, Table 3). Longitudinal observation showed that the CI-aided sound-field thresholds gradually improved after 1y in all three groups, although the improvement was not statistically significant and no intergroup differences were observed (Fig. 4b). The interaction was not significant in this analysis.

3.3. MAP parameters of the second CIs

Regarding MAP parameters of the second CI (Fig. 5), C-levels were significantly lower in group C than in groups A ($p < 0.00001$) and B ($p = 0.00014$), and in group B than in group A ($p = 0.00028$) (Fig. 5a, Table 4). T-levels were significantly lower in group C than in group A ($p = 0.03$) (Fig. 5a, Table 4). The dynamic range in group A was significantly wider than that in groups B ($p < 0.00001$) and C ($p = 0.000014$) (Fig. 5b, Table 4).

Table 4. MAP parameter values of second CI.

	C-level (Mean±SD)	T-level (Mean±SD)	Dynamic range (Mean±SD)
Group A	193.8±11.8	144.2±17.1	49.7±15.6
Group B	184.2±24.5	144.7±26.3	39.4±11.8
Group C	172.2±16.0	132.3±21.5	39.9±11.6

The mean MAP parameter values (C-level, T-level, and dynamic range) ±SD (CL: current level) of second CI at the five years after second implantation

Next, we examined the change in MAP parameters from 1 to 5 years after surgery (Fig. 5c) to evaluate if the change affects the outcome of cochlear implantation. The changes in C- and T-levels were small in group A and both C- and T-levels increased in group B. In contrast, both C- and T-levels in group C decreased from 1 to 5 years after surgery. The decrease in C- and T-levels in group C patients was significantly higher than that of the C-level in group A ($p = 0.027$) and the T-level in groups A and B ($p < 0.00001$), respectively (Fig. 5c). The change in T-level in group C was variable. Regarding the dynamic range, no appreciable change was observed in groups A and B. Group C patients showed increased dynamic range values (Fig. 5c).

4. Discussion

4.1. Effects of the second CIs

Previous studies from our group and others have shown that speech perception with the second CI alone was poorer when it was implanted at older ages or a significantly delayed timing from the first cochlear implantation [3,10]. In this study, we examined speech perception ability with the second CI alone at a much later time after the second cochlear implantation (from 3 years to 10 years) and found similar results (Figs. 1, 2) with short-term studies. As a result, speech perception with the second CI alone was significantly worse than that with bilateral CIs or the first CI alone under both quiet and noisy conditions in patients who underwent the second implantation at later than 7 years of age (Fig. 3).

Based on a previous study showing that the difference in MAP parameters affected the outcome of CI [13], we examined the MAP parameters of the second CI. Group C patients had lower C- and T-levels and a narrower dynamic range than patients who underwent bilateral cochlear implantation before the age of 3.5 years (group A patients) (Fig. 5a, b). Regarding C-levels, patients who underwent the second cochlear implantations between 3.5 and 7 years of age (group B patients) also had higher C-levels than group C patients (Fig. 5a). Accordingly, the current study suggests that poor speech perception with the second CI alone in later implanted patients was affected by MAP parameters. Considering that speech perception using the second CI alone was poorer in group C than in groups A and B in the longitudinal analysis (Fig. 2), the causative MAP parameters should differ between group C and groups A and B. Therefore, it is suggested that C-levels mostly affected speech perception with the second CI alone.

The evaluation of changes in MAP parameters of the second CI 5 years after the second CI surgery showed that, in group A, MAP parameters did not change from 1 year. C- and T-levels remained high, and the dynamic range remained wide (Fig. 5). In group B, although T-levels and the dynamic range changed slightly from 1 year, C-levels did not change at high levels (Fig. 5). These results indicate that patients who underwent second cochlear implantation before 7 years of age had stabilized and robust MAP parameters with less amount and variability of the changes. This led to better longitudinal speech perception ability in these groups when the second CI alone was used. In contrast, the second CI speech perception ability of group C patients was lower and did not improve to the level of other groups through the studied 6 years both under quiet and noise conditions (Figs. 2, 3). One of the causes of this situation, the changing MAP parameters of group C patients, is shown (Fig. 5c). In the C- and T-levels, a negative change was observed only in group C, and the dynamic range was significantly increased compared with groups A and B. Accordingly, group C patients who underwent second cochlear implantation later than 7 years of age could not obtain stabilized MAP parameters even after long-term use. These unstable MAP parameters may prevent the improvement of speech perception ability in group C. In addition, despite the increase in the dynamic range in group C over the long term, it was still narrower than that in group A (Fig. 5b) and was at the same level as the dynamic range in group B (Fig. 5b). The increase in dynamic range in group C was due to changes in T-levels (Fig. 5c), indicating difficulty in increasing C-levels. C-levels in groups A and B, which had the same dynamic range, were significantly higher than those in group C (Fig. 5a). Accordingly, to improve speech perception ability, increasing C-levels are required, and these findings are consistent with those of a previous study reporting lower C-levels of the second CI in children with longer inter-implant intervals [13,14]. These results suggest that the timing of the second cochlear implantation affects the plasticity of speech perception ability, as well as the speech perception ability itself. Moreover, the second implantation before 7 years of age is necessary to improve speech perception ability long after cochlear implantation.

In contrast to the speech perception (Fig. 2) and MAP parameters (Fig. 5a), the CI-aided sound-field thresholds of the second CI were comparable among the groups (Fig. 4a, Table 3). Additionally, CI-aided sound-field thresholds of the first and second CIs alone were not significantly different, even in group C (Fig. 4a, Table 3). These results suggest that the timing of the second cochlear implantation did not affect the sound-field thresholds of the second CI, which is different from speech perception ability.

Patients who underwent the second cochlear implantation before 7 years of age (groups A and B) seemed to be in

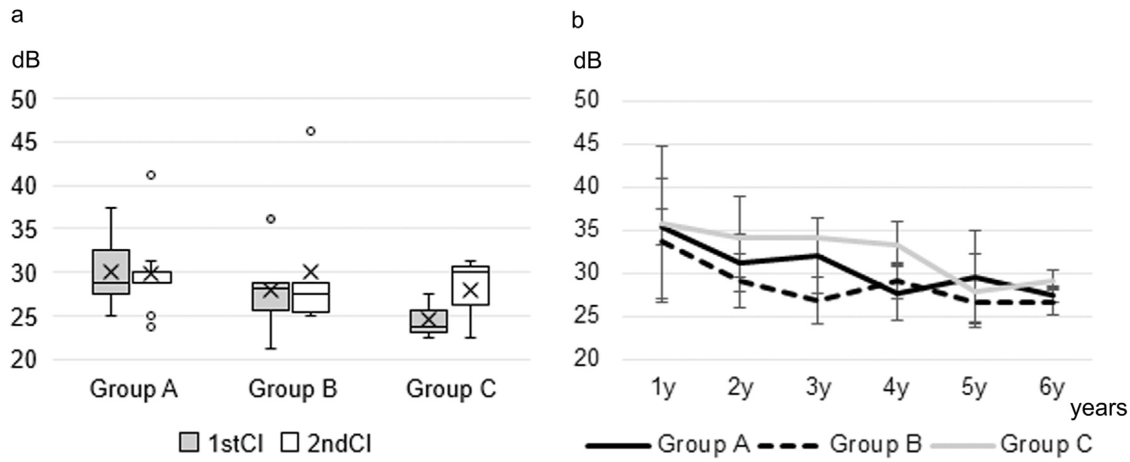

Fig. 4. Long-term outcomes of the cochlear implant-aided sound-field thresholds. Panel a shows the mean sound-field thresholds (dB HL) at the latest examination when patients wear the first or second CI alone. The details of the box plot structure are the same as those described in Fig. 1. The mean thresholds are not significantly different between using the first and second CI alone in all groups. Panel b shows the longitudinal change in the mean sound-field thresholds ± SD (dB HL) while wearing the second CI alone every year until 6 years after the second implantation. The change in mean threshold is not significantly different among each group.

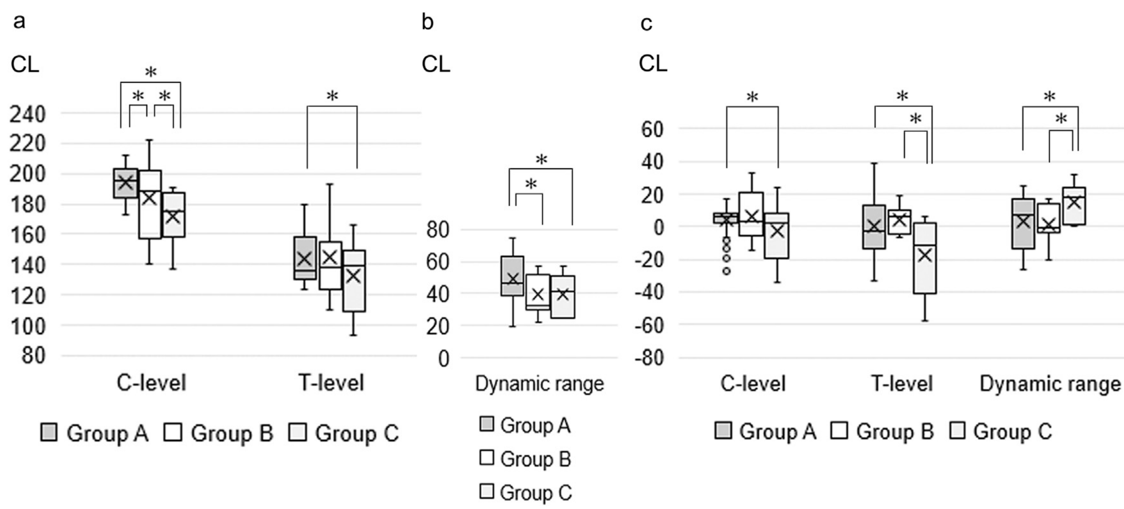

Fig. 5. MAP parameter values of the second CI. a. C- and T-levels of the second CI (CL: current level) 5 years after the second implantation. The CL for C-levels is significantly different between groups A and B, groups A and C, and groups B and C. The CL for T-levels is significantly lower in group C than in group A. b. The dynamic range of the second CI 5 years after the second implantation. The dynamic range of group A is significantly wider than that of groups B and C. c. The change in MAP parameter values of the second CIs from 1 to 5 years after the second CI implantation. Group A patients show very small changes in C- and T, and group B patients show an increase in these parameters. In contrast, patients in group C show decreased C- and T-levels. The dynamic range does not change in groups A and B but increased in group C patients. The changes in all examined parameters are significantly different between groups A and C. Groups B and C show significantly different changes in the T-level and dynamic range. * $p < 0.05$. The box plots' structure is the same as in Fig. 1.

a state of binaural balance (Figs. 1, 3) because both speech perception (Fig. 3) and CI-aided sound-field thresholds with the first and second CIs alone (Fig. 4b) were comparable. In contrast, in group C, speech perception of the second CI was significantly poor under both quiet and noisy conditions, even after long-term observation (Figs. 1–3), although the CI-aided sound-field thresholds were good (Fig. 4a). This result indicates that the effects of the second implantation are limited even after long-term observation when the second CI is implanted after 7 years of age.

4.2. *Effects of bilateral CIs*

Using bilateral CIs showed better speech perception ability than using the first CI alone after long-term longitudinal observation in groups B and C (Fig. 3), although this effect was not observed under the quiet condition. These results differ from the short-term observations in our previous study [10].

A noisy environment is a challenge for unilateral CI users. Patients who underwent second cochlear implantation aged more than 3.5 years had better speech perception ability in noise with bilateral CIs than that with the first CI (Fig. 3). This result indicated that the addition of the second CI after 3.5 years old improved the speech perception ability in noise with bilateral CIs. Our results support previous studies showing better speech perception ability in noise with bilateral CI use at 24 [15] and 36 [16,17] months after the second cochlear implantation. However, this additive effect of the second CI was observed even in group C patients who underwent the second cochlear implantation at the age of 7 years and had poor speech perception ability with the second CI. Accordingly, this effect of the second CI may be explained by the improvement of CI-aided sound-field thresholds with the second CI alone (Fig. 4).

In contrast, patients who had bilateral cochlear implantation before 3.5 years showed similar speech perception in noise between bilateral and unilateral CIs longitudinally and demonstrated a gradual increase in speech perception in noise over time. The ceiling effect may have affected the results, suggesting a need for protocol adjustments, such as the increased difficulty of presented speech or lower signal-to-noise ratio. To allow the measurement of bilateral CIs effects in young children, monosyllabic sounds were selected because of their suitability for testing and independence from language ability. However, the number of sounds was only 20 per list in the 67-S list test that we used in this study to present Japanese monosyllabic words. Future studies should consider increasing the number of sounds. Furthermore, consideration should be given to testing perception using words or sentences in children of the same age and language ability, as conducted in previous studies [18,19]. A possible study would include investigating the differences between speech perception testing using words or sentences and using monosyllabic sounds. As testing schemes, we should consider using words and sentences in tests on patients with better test results with monosyllabic sounds, or using monosyllabic sounds for younger children and words and sentences for older children. Even when testing is performed under noisy conditions resembling real-life environments, as in our previous study [10], binaural hearing benefits at present speaker positions rely on binaural summation effects. Thus, speaker positions should also be investigated to obtain advantages such as the head-shadow effect, which reportedly delivers greater binaural hearing benefits [20]. Further investigation is required regarding the optimal test conditions for measuring bilateral hearing benefits according to parameters such as time since second implantation and age of children with bilateral CIs.

5. Conclusions

When the second implantation was performed at 7 years of age or older, the second CI effects were limited, even with long-term use. The second CI-aided sound-field thresholds were good; however, speech perception when using the second CI alone remained low in patients who underwent cochlear implantation at age of >7 years.

These poor effects of the second CI may be attributed to unstable MAP parameters. When the patients underwent second cochlear implantation at age of >7 years, the MAP parameters of the second CI comprised low C- and T-levels and a narrow dynamic range even after long-term use. Among these parameters, poor speech perception may be correlated, especially with low C-levels.

Although the outcome of the second CI itself was limited, there was no difference in the CI-aided sound-field threshold between the first and second CIs alone. This contributed to the better outcome of bilateral CI use in noise, even if the second implantation was performed at 7 years of age or older.

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Author contributions

The study was conceptualized by NM and designed by NM and NY. NM, KK, and MY acquired the data. NM, NY, and TO analyzed and interpreted the data. SY provided methodological advice. JI and KO supervised the study. All authors read and approved the final manuscript.

Declaration of Competing Interest

The authors declare that there are no conflicts of interest.

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