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ORIGINAL ARTICLE

## Is there a deafness duration limit for cochlear implants in post-lingual deaf adults?

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### Abstract

**Conclusion:** Patients with post-lingual deafness should not be excluded from cochlear implantation (CI) on the basis of duration of deafness. We found that the prognosis was favorable in patients who developed deafness after adolescence, even in those with extremely long-term deafness. **Objectives:** CI is an effective treatment for post-lingual deafness. However, it remains unclear whether CI would benefit patients with extremely long-term deafness. We evaluated the auditory performance after CI of patients who had been deaf for more than 30 years. **Methods:** The study enrolled 81 adults with post-lingual deafness. Speech perception tests were performed preoperatively and 12 months postoperatively, and factors affecting the postoperative auditory performance were investigated. The subjects were divided into groups according to the duration of deafness and the postoperative speech perception scores were compared. **Results:** A marked improvement in the open-set speech perception scores (mono/disyllabic words and sentences) after implantation was seen in all groups, and no significant difference in the improvement in speech perception scores was observed among the groups. Age at onset of deafness was closely related to the postoperative performance, and patients who had lost their hearing before adolescence performed poorly.

**Keywords:** Cochlear implantation, post-lingual deafness, speech perception tests

### Introduction

Following the approval of cochlear implantation (CI) for adults with post-lingual deafness by the US Food and Drug Administration in 1984, CI has become the most effective treatment option for this condition [1]. However, it remains unclear whether CI would benefit patients with long-term hearing loss because improvement in speech recognition is related to duration of deafness [2–4].

Geier et al. [2] reported that patients who had been deaf for more than 60% of their lives exhibited slower speech recognition improvement. Roditi et al. [3] and Blamey et al. [5] both showed that improvement of auditory performance after CI is inversely proportional to duration of deafness. Ponton et al. [4] reported that prolonged deafness limits auditory

system plasticity. Post-mortem histological examinations of the temporal bones in patients with post-lingual deafness have shown that loss of inner ear hair cells induces retrograde neural degeneration, and the extent of degeneration appears to be correlated with duration of deafness [6,7].

However, most of the above studies were carried out in the 1990s, and post-CI outcomes have improved with advances in speech-processing strategies [8]. In addition, rehabilitation techniques have become more advanced, and the experience of medical professionals has improved [9]. Moreover, recently it was reported that a long duration of deafness does not always result in excessive degeneration [10]; patients with few residual spiral ganglion cells can often obtain good results [11]. Thus, further interpretation is necessary for patients with long-term hearing loss.

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We hypothesized that patients with post-lingual deafness who recently underwent CI would benefit despite a long duration of deafness. We also assumed that results from patients with long-term deafness would be similar to those of patients with short-term deafness, and that CI would result in substantial improvements in quality of life [12]. If these hypotheses are true, then age and the duration of deafness will not be limiting factors for CI. The present study evaluated CI outcomes of patients with long-term, post-lingual deafness. It also analyzed the characteristics and predictive factors of CI outcomes in these patients.

### Material and methods

Over the past two decades, 832 patients have undergone CI at our hospital. Among them, 81 adult patients who underwent surgery after 2006 and completed a follow-up speech perception test 1 year after surgery were reviewed. Patients who underwent surgery before 2006, were under age 18 at the time of surgery, had a follow-up period of less than 1 year, failed to complete the postoperative speech perception test, or exhibited prelingual hearing loss were excluded. The subject criteria for this study also included onset of deafness after age 7 to fully exclude patients with prelingual deafness [13,14]. The average age of all subjects in this study was  $54.05 \pm 14.43$  years (range 18–73 years), and the mean duration of deafness was  $17.75 \pm 13.76$  years. In all, 35 male subjects and 46 female subjects participated in the study. The causes of deafness in the participants were chronic otitis media ( $n = 9$ ), sudden hearing loss ( $n = 7$ ), trauma ( $n = 7$ ), ototoxicity ( $n = 6$ ), Meniere's disease ( $n = 2$ ), and idiopathic hearing loss ( $n = 50$ ).

Evaluations such as pure-tone audiograms (PTAs), auditory brainstem response (ABR) tests, temporal magnetic resonance imaging (MRI), and speech perception tests were performed preoperatively. PTA thresholds were examined up to 120 dB, and in cases where PTA was scaled to all frequencies, it was expressed as 120 dB HL (hearing level). Average pure-tone thresholds were calculated using results at 500, 1000, 2000, and 3000 Hz. GSI Audera (Grason-Stadler Co., Eden Prairie, MN, USA) was used for ABR tests, and the Korean version of the Central Institute of Deafness (K-CID) sentence test was used for speech perception [1]. The K-CID sentence test consisted of mono/disyllabic word and sentence perception tests under both auditory-only (AO) and auditory-visual (AV) listening conditions; tests were performed with samples of everyday words or sentences in a noiseless sound-field environment. The test was scored on percentage of words correctly repeated. The list was evaluated by two female speech

therapists, and the 'word' was checked using the adult everyday sentence with a live voice. Each set of adult everyday sentences consisted of 10 sentences including 50 words. Our institution selected one of seven types of sentence sets. AV testing was performed in full view of the patient, and AO testing was performed with the examiner's mouth hidden. Postoperative studies were conducted for CI-only and bimodal patients, and results obtained from CI-only patients were analyzed in our study. The category of auditory performance (CAP) score was measured to evaluate the patient's general auditory performance.

Patients eligible for CI were individuals who would not benefit from a hearing aid or other surgery and who had severe to profound hearing loss at the time of implantation (higher than 70 dB HL in the PTA test and 70 dB nHL in the ABR test for the ear with better hearing). In the K-CID sentence test, an AO sentence score of less than 50% was required. The implantation site was determined primarily on the possibility of bimodal stimulation after CI. If the patient exhibited preoperatively good residual hearing (>20% in best-aided status and with visual cues), used hearing aids, and had some residual hearing (<90 dB), the poorer ear was chosen for implantation.

The aetiology of deafness, average PTA thresholds in the better ear, duration of deafness, and device type were investigated. Patients had no psychomedical contraindications, and all patients were motivated for rehabilitation with realistic expectations after implantation.

After logistic regression, patients were divided into four groups according to the duration of deafness, and PTA, CAP, and K-CID sentence scores were analyzed before and after surgery using paired *t* tests and one-way analysis of variance (ANOVA). Deafness duration was defined as the difference between age at operation and age at onset of deafness. If patients showed below 20% of AO sentence scores, we grouped those as the postoperative poor auditory performance group.

In addition, factors affecting changes in postoperative K-CID sentence scores were also analyzed. Patients were categorized into groups based on factors known to have an effect on analysis, and differences in the K-CID sentence and CAP scores before and after surgery were compared using one-way ANOVA, paired *t* tests, and descriptive analysis tests. Other than duration of deafness, age at operation and age at onset of deafness also affected the results, and these variables were analyzed. Age at onset of deafness was defined as the time point that PTA threshold of the better hearing ear became worse than 70 dBHL.

The study protocol was approved by the Ethical Committee of Yonsei University College of Medicine

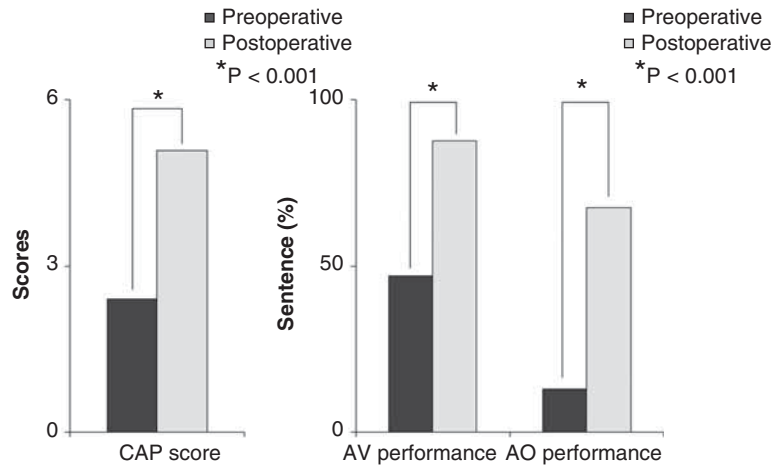


Figure 1. Overall auditory performance after cochlear implantation (CI). Average category of auditory performance (CAP), auditory-visual (AV) sentence, and auditory-only (AO) sentence scores were improved 1 year after CI.

(no. 4-2012-0474) and consents were obtained from all participants.

Data were summarized using mean and standard deviation. We used SPSS for Windows version 16.0 (SPSS Inc., Chicago, IL, USA) for the statistical analysis. A  $p$  value of  $< 0.05$  was considered statistically significant.

## Results

### Overall auditory performance after CI

In total, 77 of 81 patients displayed improved auditory performance with CI. Average CAP scores increased from  $2.40 \pm 1.86$  preoperatively to  $5.09 \pm 1.28$  postoperatively. Mean AV sentence scores increased from  $46.92 \pm 31.65$  preoperatively to  $87.63 \pm 18.68$  postoperatively. The mean preoperative AO sentence score was  $10.60 \pm 16.58$ , compared with  $46.53 \pm 27.47$  postoperatively (Figure 1). Postoperative performance for mono/disyllabic words also increased in both AV and AO listening conditions (Supplement 1).

Postoperative AV and AO performance appeared to have a moderate positive correlation with preoperative AV and AO performance ( $r = 0.250$  and  $p = 0.030$ ,  $r = 0.375$  and  $p = 0.001$ , respectively). There was no significant correlation between preoperative pure-tone threshold and postoperative AV ( $r = -0.149$ ,  $p = 0.212$ ) and AO performance ( $r = -0.159$ ,  $p = 0.177$ ).

### Predictive factors related to postoperative auditory performance

Through multiple linear regression, factors that could affect the post-surgery auditory performance were investigated, including duration of deafness, age at onset of deafness, preoperative residual hearing, and

preoperative K-CID scores. We found that age at implantation, duration of deafness, and age at onset of deafness all affected post-surgical outcomes (Table I). The cause of deafness, device type, speech-processing strategy, education level, and other various causes had no significant impact on post-implantation auditory outcomes (data not shown). Postoperative K-CID scores for all subjects were compared and analyzed after categorization into factor-based groups.

### Differences according to the duration of deafness

We categorized all patients into four groups based on duration of deafness, and no differences in gender, age at operation, preoperative hearing threshold, preoperative auditory performance, cause of hearing loss, or general condition were identified. However, a difference in the age at onset of deafness was noted (Table II). In all groups, auditory performance improved after surgery (Supplement 2). In analyses of postoperative auditory performance, no significant differences in AV performance were detected among the four groups but significant differences in postoperative AO performance were identified ( $p < 0.05$ ; Figure 2A and Supplement 3). No differences were observed (Figure 2B and Supplement 3) in the degree of improvement in auditory performance among the groups (preoperative and postoperative score difference).

### Effect of age at implantation

Age at implantation affected postoperative auditory performance. After categorizing patients into good and poor responding groups based on postoperative auditory performance, logistic regression was performed to specify the age at which differences were

Table I. Predictive factors related to postoperative auditory performance.

Factor	Postoperative AO performance		Postoperative AV performance		Postoperative CAP score	
	$\beta$	<i>p</i> Value	$\beta$	<i>p</i> Value	$\beta$	<i>p</i> Value
Age at implantation (years)	1.80	0.025*	0.95	0.030*	-0.23	0.190
Age at onset of deafness (years)	-2.09	0.012*	-1.18	0.012*	0.116	0.001*
Duration of deafness (years)	-2.95	0.020*	-1.53	0.020*	0.20	0.155
Preoperative PTA threshold (dB HL)	-0.31	0.344	-0.33	0.067	-0.81	0.580
Preoperative SDS (%)	0.14	0.257	0.098	0.216	0.27	0.643
Preoperative CAP score	-1.97	0.594	-4.65	0.045	-0.19	0.897
Preoperative K-CID AV sentence score	0.36	0.101	0.17	0.154	0.03	0.962
Preoperative K-CID AO sentence score	0.98	0.723	-0.10	0.686	-0.32	0.808

Through multiple linear regressions, it has been determined that age at operation, deaf duration, and age at deafness are the meaningful factors. AO, auditory-only listening condition; AV, auditory-visual listening condition; CAP, category of auditory performance; K-CID, Korean version of the Central Institute of Deafness (K-CID) sentence test; PTA, pure-tone audiogram; SDS, speech discrimination score.

\*Statistically significant.

observed. The cut-off point on the receiver operating characteristics (ROC) curve was set at 60 years old (data not shown).

When we analyzed the patients younger ( $n = 45$ ) and older ( $n = 36$ ) than 60 years, we observed no significant difference in AO results after surgery (Figure 3B).

#### *Differences according to preoperative K-CID scores and preoperative residual hearing*

According to multiple linear regression analysis, preoperative auditory performance may not affect CI outcomes. Using linear logistic regression after

categorizing patients into good and poor responding groups based on postoperative auditory performance (see Analysis of the characteristics of the poor performance group, below), the authors did not establish cut-off points on the ROC curve for preoperative AV and AO sentence scores (data not shown).

Similarly, preoperative residual hearing of the implanted ear may not affect surgical outcomes. However, we compared postoperative AO and AV sentence scores for degree of residual hearing in the better ear and divided patients into two groups: severe hearing loss ( $n = 25$ ) and profound hearing loss ( $n = 56$ ). Contrary to our expectation, there was no significant difference between the groups (Figure 3A).

Table II. Comparison of preoperative characteristics among groups divided according to duration of deafness.

Characteristic	$\leq 10$ years ( $n = 24$ )	10–20 years ( $n = 26$ )	20–30 years ( $n = 14$ )	$> 30$ years ( $n = 17$ )	<i>p</i> Value
Sex (male:female)	20:13	20:14	9:10	10:13	0.530
Age at implantation (years)	53.09 $\pm$ 17.66	57.58 $\pm$ 15.22	48.72 $\pm$ 13.35	56.08 $\pm$ 8.06	0.182
Age at onset of deafness (years)	48.06 $\pm$ 17.59	44.26 $\pm$ 15.20	24.89 $\pm$ 14.78	16.39 $\pm$ 7.49	$< 0.001^*$
Duration of deafness (years)	4.42 $\pm$ 2.59	13.23 $\pm$ 3.16	22.72 $\pm$ 3.77	39.69 $\pm$ 7.11	$< 0.001^*$
Preoperative PTA threshold* (dB HL)	98.2 $\pm$ 17.50	100.03 $\pm$ 14.03	106.29 $\pm$ 10.75	94.43 $\pm$ 23.06	0.189
Preoperative SDS† (%)	2.47 $\pm$ 13.51	23.65 $\pm$ 39.93	5.18 $\pm$ 21.34	0.00 $\pm$ 0.00	0.002
Preoperative CAP score	2.48 $\pm$ 2.26	2.78 $\pm$ 1.41	2.00 $\pm$ 1.89	2.61 $\pm$ 1.53	0.565
Preoperative K-CID AV sentence score	39.27 $\pm$ 35.25	51.00 $\pm$ 34.00	48.62 $\pm$ 25.22	54.38 $\pm$ 31.02	0.384
Preoperative K-CID AO sentence score	20.97 $\pm$ 26.57	12.68 $\pm$ 16.63	10.67 $\pm$ 17.48	9.52 $\pm$ 17.05	0.196
Onset of deafness $< 13$ years	3/24	2/26	3/14	10/17	0.001*
Age at implantation $> 60$ years	10/24	16/26	5/14	5/17	0.169

AO, auditory-only listening condition; AV, auditory-visual listening condition; CAP, category of auditory performance; K-CID, Korean version of the Central Institute of Deafness (K-CID) sentence test; PTA, pure-tone audiogram; SDS, speech discrimination score.

\*PTA threshold of implanted ear.

†SDS of implanted ear.

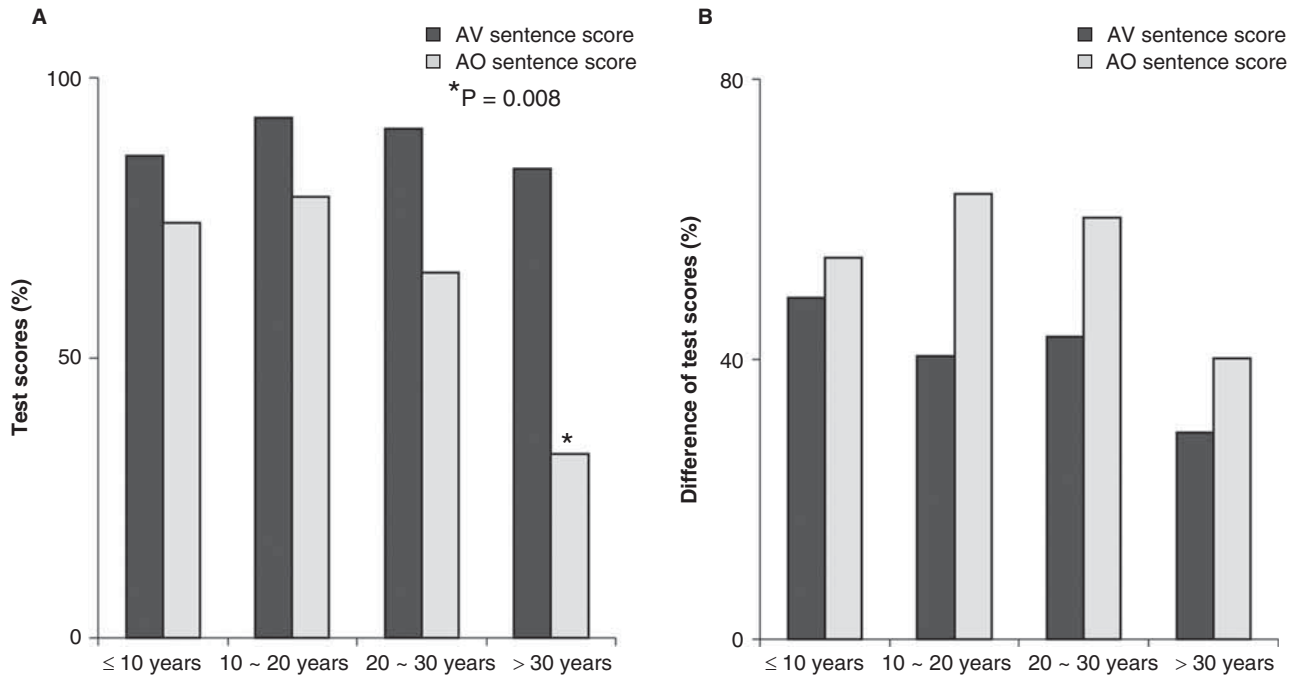


Figure 2. Comparison of auditory performance based on the duration of deafness. (A) When we compared postoperative auditory performance based on duration of deafness, there was no significant difference in auditory-visual (AV) sentence scores between groups, but a significant difference in auditory-only (AO) sentence scores was noted at more than 30 years duration of deafness ( $p = 0.008$ ). (B) When comparing the degree of improvement in auditory performance between groups (preoperative and postoperative score difference), no differences were detected for AV and AO sentence score improvement.

*Differences according to the age at onset of deafness*

Age at onset of deafness affected postoperative auditory performance. After categorizing patients into good and

poor responding groups based on postoperative auditory performance, logistic regression was performed to specify the age at which differences were observed. Most patients developed hearing loss at age 13. Then

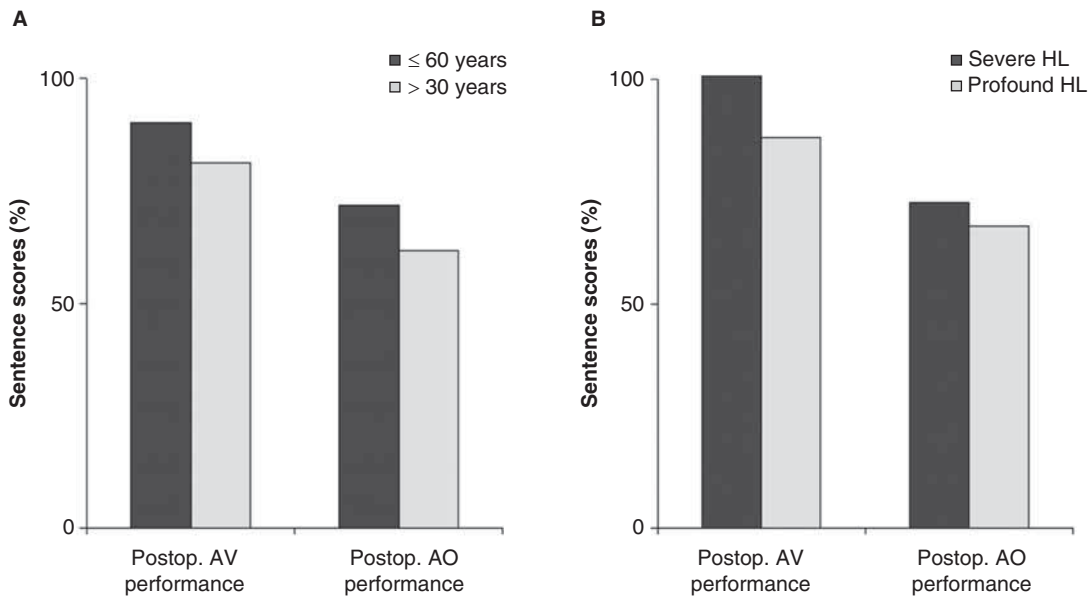


Figure 3. Comparison of auditory performance according to other factors. (A) Patients were divided based on an implantation age of 60 years, and no significant differences in postoperative AV and AO sentence scores were observed ( $p = 0.063$  and  $0.152$ , respectively). (B) Preoperative residual hearing may not affect cochlear implantation (CI) outcomes. No significant differences were observed in postoperative auditory-visual (AV) and auditory-only (AO) sentence scores ( $p = 0.139$  and  $0.512$ , respectively) between severe and profound hearing loss groups.

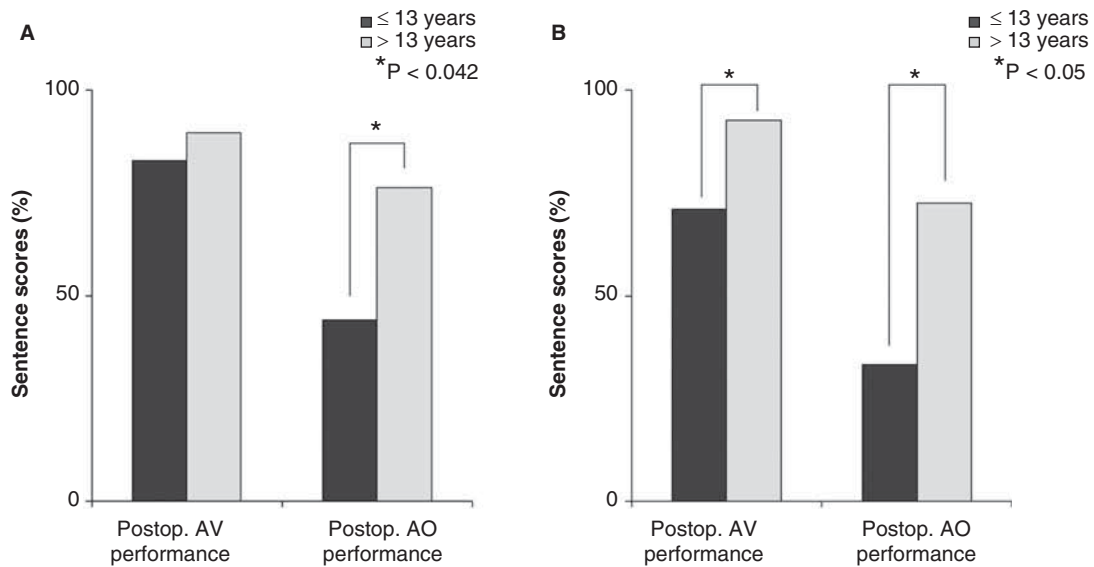


Figure 4. Comparison of auditory performance based on age at onset of deafness. (A) Patients were divided based on age at onset of deafness of greater or less than 13 years, and a significant difference in postoperative auditory-only (AO) sentence scores was observed ( $p = 0.042$ ). (B) In the long-term deafness group ( $>30$  years), outcomes for people who lost their hearing after age 13 were superior to those of patients who lost their hearing before age 13. Significant differences in both auditory-visual (AV) ( $p = 0.039$ ) and AO ( $p = 0.001$ ) sentence scores were detected.

when we analyzed the patients after dividing them into two groups, younger ( $n = 18$ ) and older ( $n = 63$ ) than 13 years, we found a significant difference in AO results after surgery ( $p = 0.042$ , Figure 4A).

#### *Analysis of the characteristics of the poor performance group*

The patients in our study were easily separated into groups based on postoperative auditory performance. A total of 16 patients had poor postoperative AO sentence scores (below 20%, poor performance group), and 48 patients had scores exceeding 50% (good performance group). We compared patient characteristics between the two groups and, contrary to our expectations, there was no significant difference in preoperative auditory performance (0% vs 11.2%,  $p = 0.287$ ). Residual hearing was significantly lower in the poor performance group (102 dB vs 89 dB,  $p < 0.05$ ), and we determined that a higher proportion of the poor performance group had an earlier onset of deafness ( $<13$  years) (11/16, 68.8%) than in the good performance group (2/48, 4.2%;  $p < 0.001$ ).

#### *Analysis of patients with long-term deafness*

Contrary to our initial expectations, patients with a duration of deafness exceeding 30 years displayed significantly higher postoperative scores for PTA (preoperative vs aided), Speech audiograms (preoperative vs aided), CAP, AO mono/disyllabic and sentence, and AV mono/disyllabic and sentence yield tests

compared with preoperative scores ( $p < 0.05$ ; Figure 1 and Supplement 1). When analyzing factors affecting the outcomes of patients with a duration of deafness exceeding 30 years, age at onset of deafness was identified as the only important factor. In addition, outcomes for people with hearing loss onset after age 13 were superior to those of patients with hearing loss before age 13 (Figure 4B and Supplement 4). The long-term deafness group, which included patients with durations of deafness greater than 30 years, exhibited a significantly higher ratio of patients whose age at onset of deafness was younger than age 13 compared with other groups ( $p < 0.001$ ; Table II). When postoperative auditory performance was examined only in the group whose age at onset of deafness was over 13 years, improvement was clearly observed. In addition, the postoperative AO score was not significantly lower in the extremely long-term deafness group compared to the scores of other groups (Supplement 5).

## **Discussion**

Many previous studies have reported that duration of deafness, age at implantation, and preoperative residual hearing are important factors for postoperative auditory outcomes in adults with post-lingual deafness [3,12,13]. However, those factors are correlated and can influence postoperative improvements in combination. Thus, it is important to choose the factor with the largest effect after considering their correlation. In the present study, using a multiple

regression test to analyze this correlation, we found that the duration of deafness, age at implantation, and age at onset of deafness influenced postoperative outcomes. In addition, the age at onset of deafness was the most critical factor for predicting post-CI auditory performance.

When analysis was based on the duration of deafness, the most important finding was that patients in all groups exhibited improved auditory performance after CI regardless of duration of deafness. The auditory improvement of the long-term hearing loss group was compared to improvement in the short-term hearing loss group (Figure 2B). This improvement in all patients is believed to be due to the fact that all patients received some degree of auditory stimulation using hearing aids or other devices regardless of the duration of deafness.

Ordinarily, the age at which an operation is performed is closely related to the patient's cognitive ability. Thus, patient age adversely affects the progress of rehabilitation after surgery (i.e. postoperative results). Previous studies [14,15] have reported that age at implantation has the greatest effect on postoperative outcomes. Nevertheless, when analyzed using 60 years as a cut-off age for logistic regression, the postoperative outcomes were not noticeably different (Figure 3B). Moreover, no differences were found between the long- and short-term deafness groups regarding the age at implantation and the percentage of patients older than 60 years (Table II). Therefore, age at implantation is not a critical factor for outcomes; this result stems from the fact that duration of deafness is not related to age at implantation but rather to the age at onset of deafness.

From the results, age at onset of deafness has a greater impact on postoperative auditory performance. We observed poor outcomes in patients who developed deafness before adolescence. This finding suggests that if hearing deteriorates before the critical period of language acquisition, it could result in poor postoperative outcomes. Patients who had been deaf for more than 30 years were more affected by age at onset of deafness. The proportion of patients who became deaf before the age of 13 (58.9%) was significantly higher than the proportion of that in other groups (7.7–21.4%). This finding may explain why patients who had been deaf for more than 30 years had lower AO performance scores. When all postoperative auditory performance scores for each group of patients who became deaf after age 13 were compared, the differences among AO sentence scores were small (Supplement 5). The same was true when considering language production: the production of spoken or written language was not exactly correlated with the duration of deafness or

current residual hearing. That of patients who became deaf at adolescence was far superior to that of patients who became deaf after adolescence.

Our results suggest that, even with long-term deafness, patients who acquired language and completed language development before age at onset of deafness constantly received linguistic neural stimulation. Therefore, hearing, as opposed to speaking, reading, or thinking using the mother-tongue language, should be their only impediment. Nevertheless, subjects with under-developed linguistic capabilities display ignorance more often despite a shorter duration of deafness because their recall of linguistic information may be limited. This theory appears to be correlated with the finding that people who have spent most of their life abroad after adolescence almost forget their native language, but after little training, they successfully re-acquire it [15–18].

The age that distinguishes prelingual and post-lingual deafness has yet to be clarified, and researchers have reported various ages ranging from 7 to 12 years as the standard. In this study, we included patients with a minimum age at onset of deafness of 7 to exclude patients with prelingual deafness. Considering that most patients in the poor performance group became deaf before age 13, age at onset of deafness clearly had the strongest influence on study outcomes. Therefore, ages 7–12 years should be considered an incomplete post-lingual stage, during which mature language has not been achieved despite language acquisition [18–20]. For patients in this category, the postoperative prediction or decision for implant must not be approached in the same manner as that for the typical patient with post-lingual deafness. Therefore, when future CI is performed for post-lingual deafness, age at onset of deafness should be considered to be a critical factor.

## Conclusion

In adults with post-lingual deafness, duration of deafness should not be a limit for cochlear implants. Age at onset of deafness is a more critical factor than duration of deafness.

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## Supplementary material available online

Supplements 1–5