on the visual and visuospatial processing subserved by the early visual cortex and parietal cortices.[33] In our study, however, hypometabolism was observed in the visual-related regions of the posterior medial lingual cortex (BA18 and BA19). We speculate that this result may be related to the fact that both subjects had received intensive auditory-verbal/oral education without visual sensation since childhood. Because communication without visual information is a part of the auditory-verbal/oral education, it may be thought that the auditory center of a subject who was educated in this manner becomes active so as to avoid utilizing visual pathways during listening and that hypometabolism in the visual center thus develops.

In this study, we used the iNEUROSTAT+ program for anatomic standardization and 3-dimensional Stereotactic Surface Projection (3D-SSP), which can more correctly translate data from an an atrophied brain to that of a standard brain[18] and complement inappropriate anatomic standardization.[17] Moreover, Statistical Parametric Mapping (SPM) normalizes the metabolic counts to global counts, and iNEUROSTAT+ can normalize pixel values of an individual's image set to the whole brain, the thalamus, the pons, the cerebellum, or the sensorimotor region and select a suitable region as a reference region for normalization.[19]

CONCLUSIONS

Even in prelingually deafened adults, intensive training by auditory-verbal/oral communication since childhood may activate the auditory-related cortices by language stimulation through a CI as in postlingually deafened successful CI users, and might result in exceptionally good CI performance, as in the present 2 subjects. Despite the limits imposed by the small sample size and the spatial resolution of PET, this study yields insights into the nature of the brain plasticity in successful adult users of CIs with prelingual deafness.

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Original Article

Bilateral Cochlear Implantation for Children in Nagasaki, Japan

Yukihiko Kanda, MD[1,2] · Hidetaka Kumagami, MD[2] · Minoru Hara, MD[2] · Yuzuru Sainoo, MD[2] · Chisei Sato, MD[2]
Tomomi Yamamoto-Fukuda, MD[2] · Haruo Yoshida, MD[2] · Akiko Ito[1] · Chiharu Tanaka[1] · Kyoko Baba[1] · Ayaka Nakata[1]
Hideo Tanaka[1] · Haruo Takahashi, MD[2]

[1]*Kanda ENT Clinic, Nagasaki Bell Hearing Center, Nagasaki;* [2]*Department of Otolaryngology Head and Neck Surgery, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan*

Objectives. The number of patients with bilateral cochlear implant (CI) has gradually increased as patients and/or parents recognize its effectiveness. The purpose of this report is to evaluate the efficacy of 29 bilateral CI out of 169 pediatric CI users, who received auditory-verbal/oral habilitation at our hearing center.

Methods. We evaluated the audiological abilities 29 Japanese children with bilateral CIs including wearing threshold, word recognition score, speech discrimination score at 1 m from front speaker (SP), 1 m from second CI side SP, speech discrimination score under the noise (S/N ratio=80 dB sound pressure level [SPL]/70 dB SPL, 10 dB) at 1 m from front SP, word recognition score under the noise (S/N ratio=80 dB SPL/70 dB SPL, 10 dB) at 1 m from front SP.

Results. Binaural hearing using bilateral CI is better than first CI in all speech understanding tests. Especially, there were significant differences between the results of first CI and bilateral CI on SDS at 70 dB SPL ($P=0.02$), SDS at 1 m from second CI side SP at 60 dB SPL ($P=0.02$), word recognition score (WRS) at 1 m from second CI side SP at 60 dB SPL ($P=0.02$), speech discrimination score (SDS) at 1 m from front SP under the noise (S/N=80/70; $P=0.01$) and WRS at 1 m from front SP under the noise (S/N=80/70; $P=0.002$). At every age, a second CI is very effective. However, the results of under 9 years old were better than of over 9 years old on the mean SDS under the noise (S/N=80/70) on second CI ($P=0.04$). About use of a hearing aid (HA) in their opposite side of first CI, on the WRS and SDS under the noise, there were significant differences between the group of over 3 years and the group of under 10 months of HA non user before second CI.

Conclusion. These results may show important binaural effectiveness such as binaural summation and head shadow effect. Bilateral CI is very useful medical intervention for many children with severe-to-profound hearing loss in Japan as well as elsewhere.

Key Words: *Cochlear implant, Children, Bilateral, Binaural, Binaural summation, Head shadow effect, Japan*

INTRODUCTION

The clinical effects of both unilateral and bilateral cochlear implantation (CI) in children is well established internationally (1-15), but there are only a few reported cases of bilateral CI in Japan. The number of patients with bilateral CI has gradually increased as patients and/or parents recognize its effectiveness. The following are some of the bilateral CI cases in children that we have experienced. This is a review of bilateral CI in 169 pediatric CI users, who received auditory-verbal/oral habilitation at our clinic.

This study's aim is to obtain answers to the following questions. 1) Until what age the second CI is effective for better language perception in various situations? 2) Does the use of a hearing aid (HA) on the opposite side of first CI affect the re-

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• Corresponding author: **Yukihiko Kanda, MD**
Kanda ENT Clinic, Nagasaki Bell Hearing Center, Wakakusa 4-25, Nagasaki 852-8023, Japan
Tel: +81-95-841-7038, Fax: +81-95-841-7041
E-mail: n-bell@estate.ocn.ne.jp

sults after second CI? 3) Is there any critical time span between the first and second CI for their progress in language perception? 4) What is the advantage of bilateral CI over unilateral CI?

MATERIALS AND METHODS

Subjects

Since we started CI surgery in 1997, out of 169 children undergoing CI rehabilitation in our clinic, 29 children (17%) had bilateral CI for at least half a year before May 2011. The age of the children at the first CI operation ranged from 1 year 4 months to 15 years 5 months, whereas the age of children at the second CI operation ranged from 2 year 1 month to 15 years 10 months (Fig. 1). The most common age for the first CI was 1 or 2 years. The interval between first and second CI fitting ranged from 5 months to 10 years 1 month. This can be considered a relatively wide range, but the most frequent interval between the two CIs was under 1 year (Fig. 2). The period on non-use of their HA before the second CI is also valuable: it ranges from 0 month to

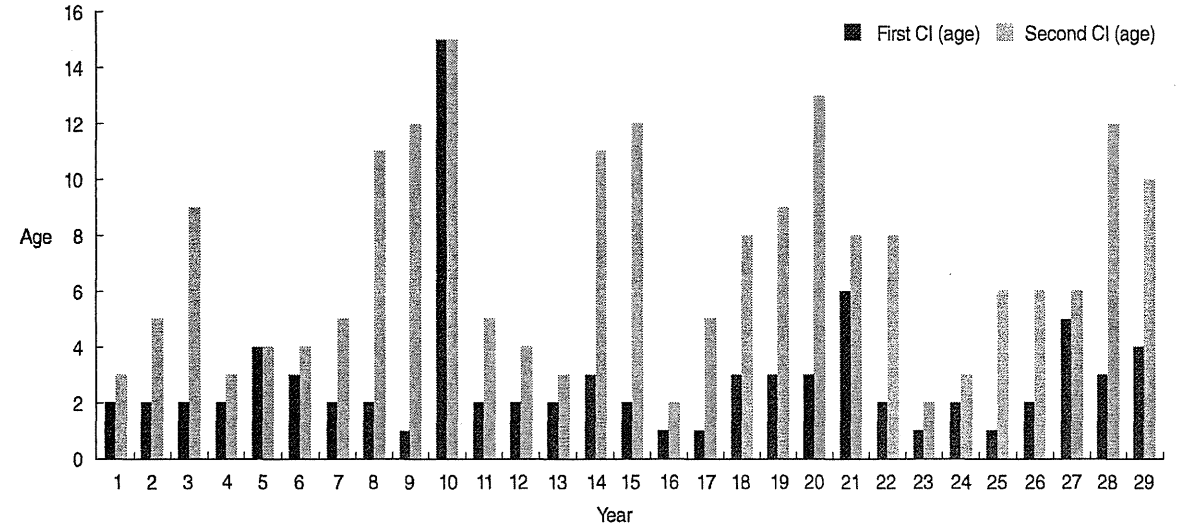


Fig. 1. Age at operation of first cochlear implantation (CI) and second CI (year).

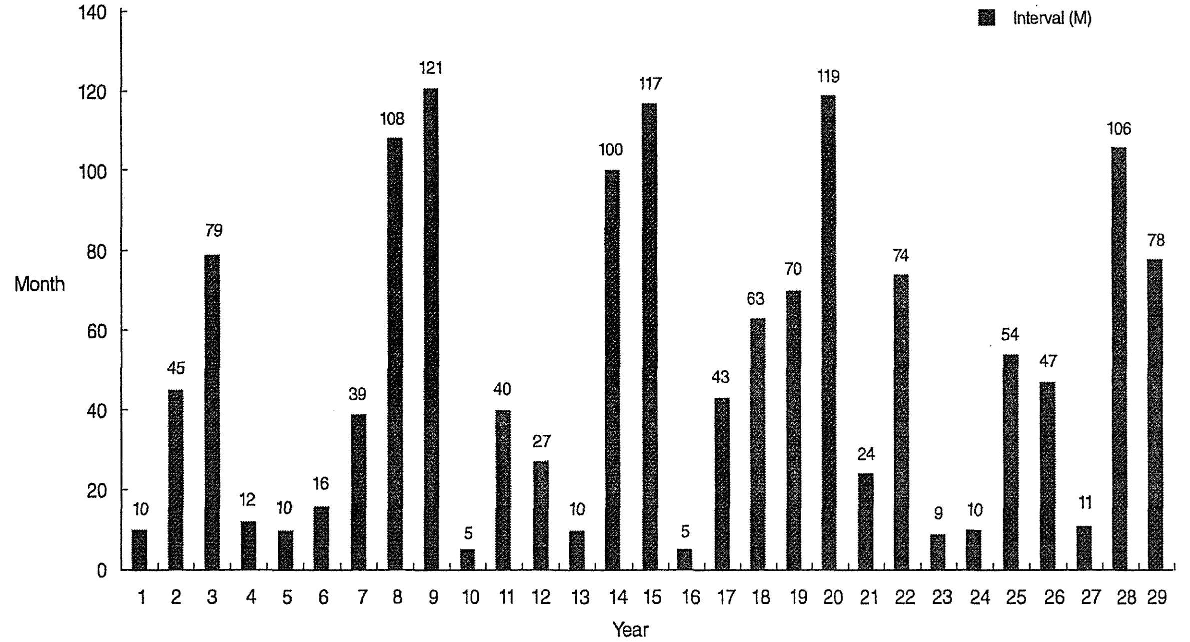


Fig. 2. Interval between first and second cochlear implantation (month).

108 months (9 years). Twelve cases did not remove their HA before the second CI (Fig. 3). Causes for deafness were described in Fig. 4. The devices used are described in Fig. 5.

We examined 19 children who acquired language with either CI or HA using various audiological tests. Children with severe anomaly or late development were not included. The children were divided into 2 groups to evaluate the amount of habilitation time after the second CI. The first group (group A) consisted of 11 children who had their second CI for at least a year. The second group (group B) included 8 children who had their second CI between 6 and 12 months.

Methods

We evaluated audiological abilities including 1) wearing threshold (WTH); 2) word recognition score (WRS, TY-89; Japanese-3 syllabic word-CD, at 60 dB sound pressure level [SPL], at 70 dB SPL); 3) speech discrimination score (SDS, 67-S; Japanese-monosyllabic word-CD, at 60 dB SPL, at 70 dB SPL) at 1 m from front speaker (SP), 1 m from second CI side SP; 4) SDS under noise (67-S; Japanese-monosyllabic word-CD, S/N ratio=80 dB SPL/70 dB SPL, 10 dB) at 1 m from front SP; 5) WRS under noise (TY-89; Japanese-3 syllabic word-CD, S/N ratio=80 dB SPL/70 dB SPL, 10 dB) at 1 m from front SP (noise: speech noise). We conducted all tests in a shielded room. Statistical analysis was done using the Student's *t*-test and paired *t*-test.

RESULTS

The mean WTH using first CI, second CI, and bilateral CI shows that all WTH is nearly the same ranging from 25 dB hearing level (HL) to 35 dB HL (Fig. 6). There were no significant differences between them. The mean WTH of their HA before he second CI was from 55 dB HL (for lower frequencies) to 65 dB HL (for higher frequencies). However, after operation the mean WTH using second CI ranges from almost 30 dB HL to 35 dB HL. There were significant difference (P=0.03*) between HA and

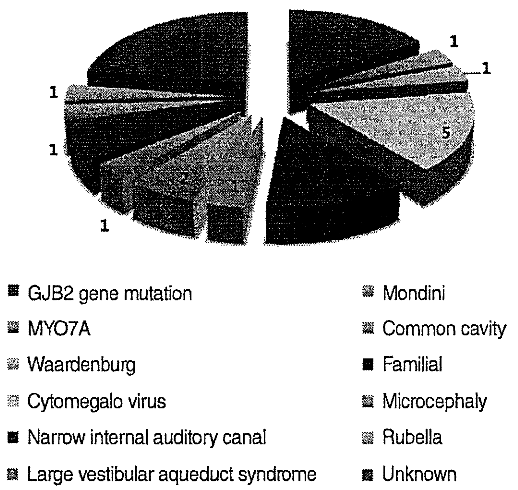


Fig. 4. Causes for deafness.

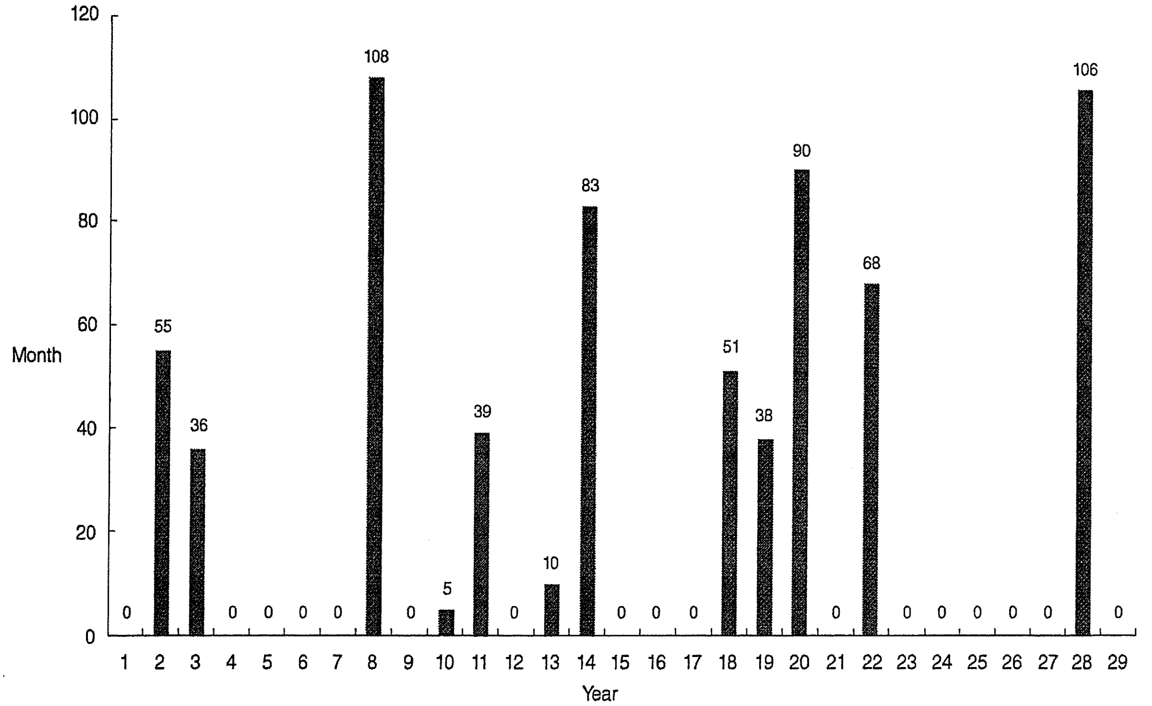


Fig. 3. Number of months child discontinued hearing aid use before second cochlear implantation.

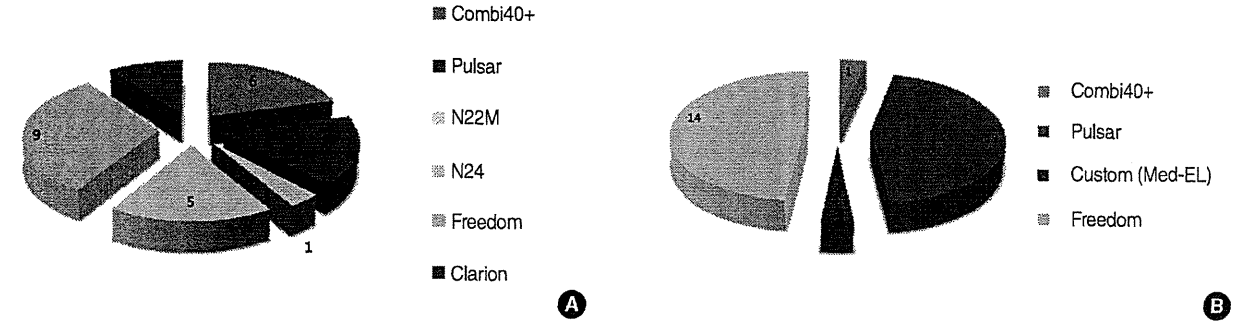
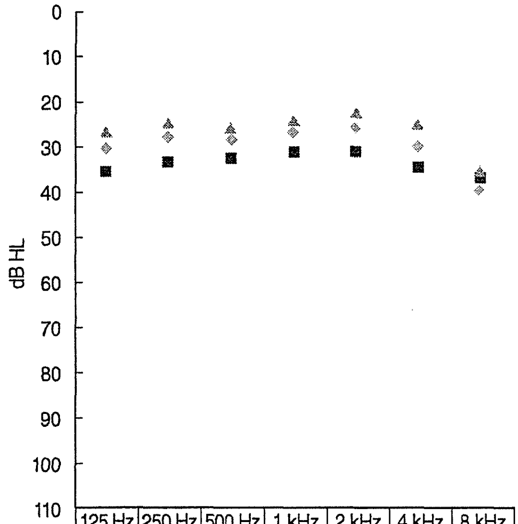
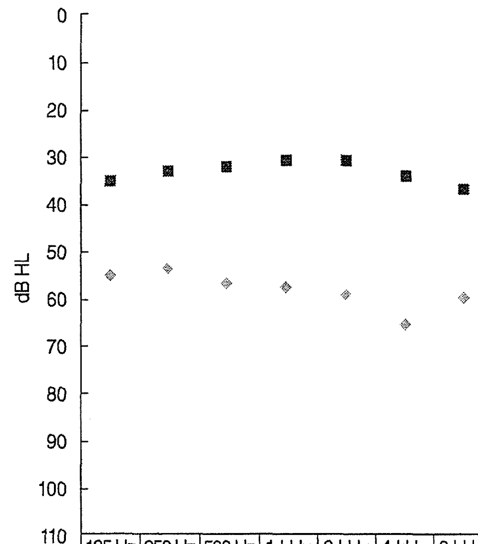


Fig. 5. Device of first cochlear implantation (CI) (A) and second CI (B).



	125 Hz	250 Hz	500 Hz	1 kHz	2 kHz	4 kHz	8 kHz
◈ First CI	30.3	27.8	28.3	26.8	25.8	29.8	39.5
■ Second CI	35.5	33.3	32.5	31	31	34.3	37
▲ Bilateral CI	26.8	24.8	25.8	24.3	22.5	25	35.4

Fig. 6. The mean wearing threshold using first cochlear implantation (CI), second CI, and bilateral CI.



	125 Hz	250 Hz	500 Hz	1 kHz	2 kHz	4 kHz	8 kHz
◈ HA	55.4	54.1	57	57.9	59.4	65.8	60
■ Second CI	35.5	33.3	32.5	31	31	34.3	37

Fig. 7. The mean wearing threshold of their hearing aid (HA) before second cochlear implantation (CI) and second CI.

second CI (Fig. 7).

The mean WRS at 1 m from the front SP at 70 dB SPL is shown in Fig. 8. The mean score for the second CI in group A was similar to the mean score for the first CI. The mean score on the WRS for all cases shows that there were no significant differences between the results of the first CI and the bilateral CI at 70 dB SPL (P= 0.13).

The mean WRS at 1 m from the front SP at 60 dB SPL is described in Fig. 9. The mean score for the second CI in group A was similar to the mean score for the first CI. For all cases, there were no significant differences between the results of the first CI and bilateral CI at 60 dB SPL (P=0.05).

The mean SDS at 1 m from the front SP at 70 dB SPL is described in Fig. 10. The mean score for the second CI in group A was similar to the mean score for the first CI. The SDS results show that there were significant differences between the results of the first CI and the bilateral CI at 70 dB SPL (P=0.02*).

The mean SDS at 1 m from the front SP at 60 dB SPL is described in Fig. 11. There were no significant differences between the results of the first CI and the bilateral CI at 60 dB SPL (P=0.24).

The mean SDS at 1 m from the second CI side SP at 70 dB SPL is described in Fig. 12. The mean score for the second CI in group A was similar to the mean score for the first CI. There were no significant differences between the results of the first CI and the bilateral CI at 1 m from the second CI side SP on all cases at 70 dB SPL (P=0.25).

The mean SDS at 1 m from the second CI side SP at 60 dB SPL is described in Fig. 13. The mean score for second CI in group A was superior to the mean score for first CI. There were significant differences between the results of first CI and bilateral CI at 1 m from second CI side SP on all cases at 60 dB SPL (P=0.02*).

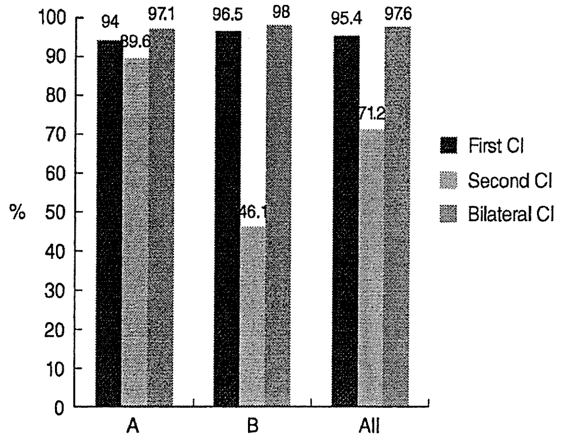


Fig. 8. Mean word recognition score at 1 m from front speaker at 70 dB SPL (0.13). CI, cochlear implantation; SPL, sound pressure level.

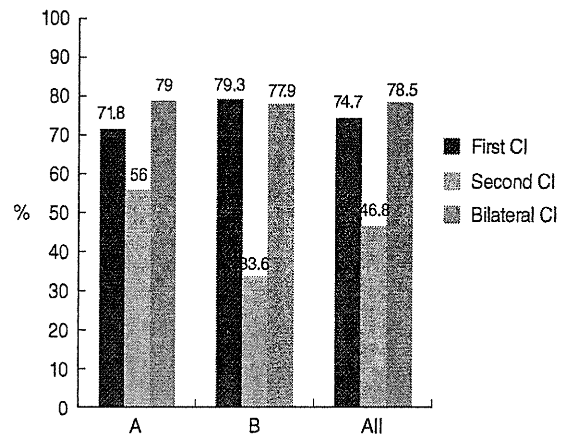


Fig. 11. Mean speech discrimination score at 60 dB SPL (P=0.24). CI, cochlear implantation; SPL, sound pressure level.

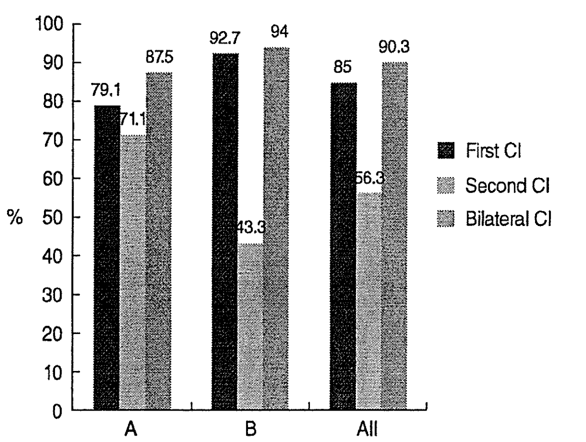


Fig. 9. Mean word recognition score at 1 m from front speaker at 60 dB SPL (P=0.05). CI, cochlear implantation; SPL, sound pressure level.

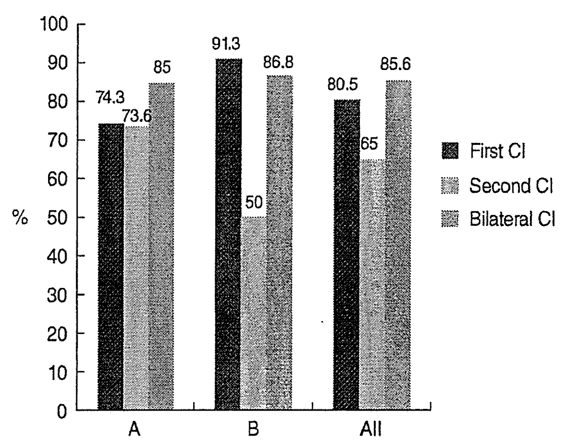


Fig. 12. Mean speech discrimination score at 1 m from second cochlear implantation (CI) side speaker at 70 dB SPL (P=0.25). SPL, sound pressure level.

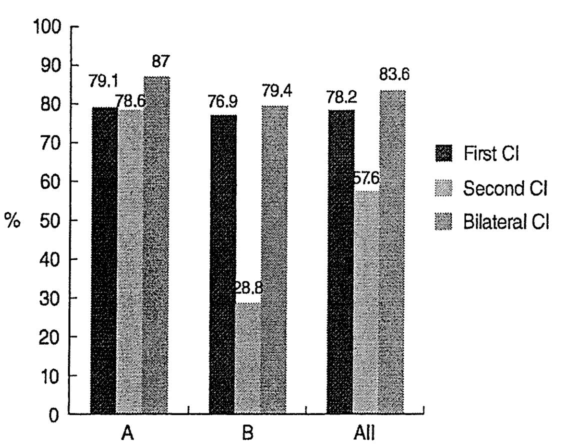


Fig. 10. Mean speech discrimination score at 70 dB SPL (P=0.02*). CI, cochlear implantation; SPL, sound pressure level.

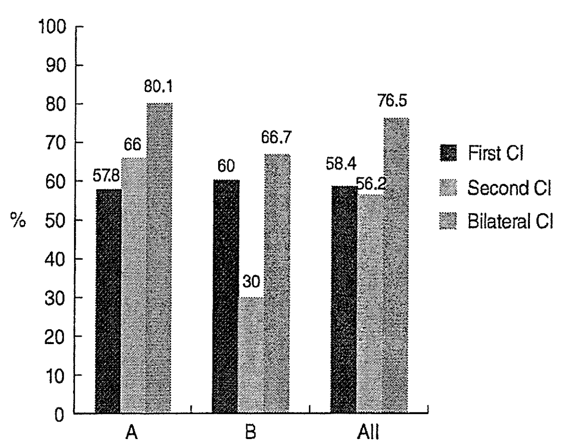


Fig. 13. Mean speech discrimination score at 1 m from second cochlear implantation (CI) side speaker at 60 dB SPL (P=0.02*). SPL, sound pressure level.

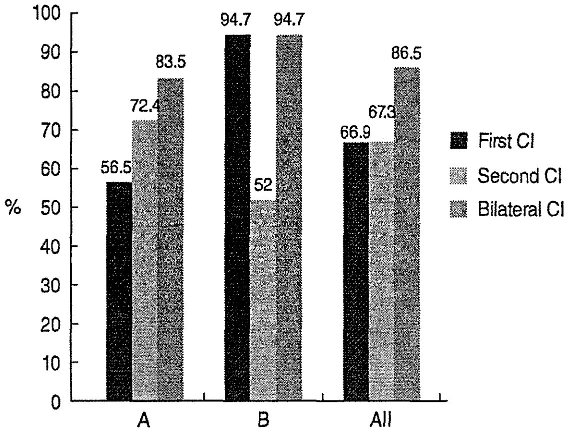


Fig. 14. Mean word recognition score at 1 m from second cochlear implantation (CI) side speaker at 60 dB SPL (P=0.02*). SPL, sound pressure level.

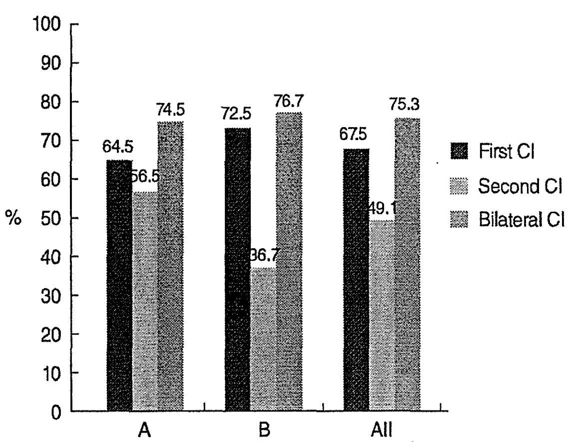


Fig. 15. Mean speech discrimination score at 1 m from front speaker under the noise (S/N=80/70, +10; P=0.01*). CI, cochlear implantation.

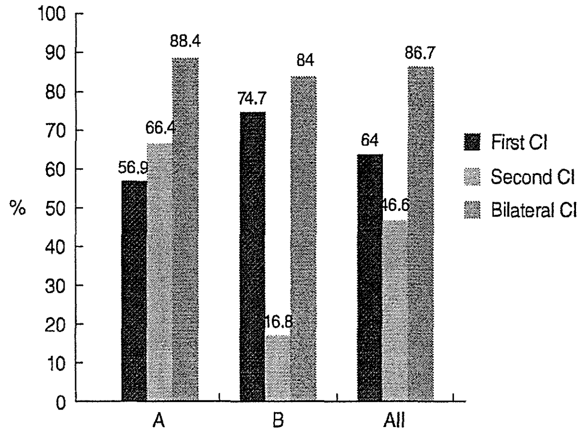


Fig. 16. Mean word recognition score at 1 m from front speaker under the noise (S/N=80/70, +10; (P=0.002**). CI, cochlear implantation.

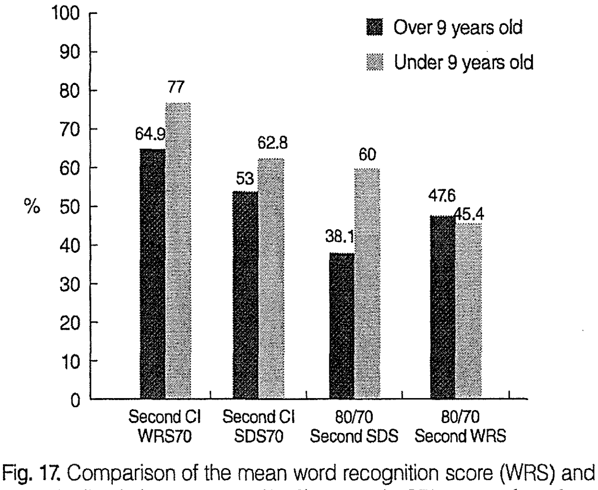


Fig. 17. Comparison of the mean word recognition score (WRS) and speech discrimination score (SDS), at 70 dB SPL at 1 m from front speaker, the mean SDS and WRS under the noise (S/N=80/70) on second cochlear implantation for over and under 9 years old. SPL, sound pressure level.

The mean WRS at 1 m from second CI side SP at 60 dB SPL were described in Fig. 14. The mean score for second CI in group A was superior to the mean score for first CI. There were significant differences between the results of first CI and bilateral CI at 1 m from second CI side SP on all cases at 60 dB SPL (P=0.02*).

The mean SDS at 1 m from front SP under the noise (S/N=80/70, +10) were described in Fig. 15. The mean score for second CI in group A was similar to the one for first CI. There were significant differences between the results of first CI and bilateral CI under the noise at 1 m from front SP on all cases (P=0.01*). The mean WRS at 1 m from front SP under the noise (S/N=80/70, +10) were described in Fig. 16. The mean score for second CI in group A was superior to the mean score for first CI. In all cases, there were significant differences between the results of first CI and bilateral CI under the noise at 1 m from front SP (P=0.002**).

We attempted to determine until what age the second CI is effective for better language perception in various situations. We compared the results of over 9 years old with the results of under 9 years old, analyzing the mean WRS and SDS at 70 dB SPL at 1 m from the front SP, the mean SDS and WRS under the noise (S/N=80/70) on second CI (Fig. 17). The mean SDS under the noise (S/N=80/70) for the second CI (P=0.04*) shows significant differences between the over 9 years old and the under 9 years old.

We compared children that had not used their HA for over 3 years before the second CI with those that had used their HA within 10 months before the second CI using various speech understanding tests (Fig. 18). The mean WRS and SDS revealed better scores for HA usage within 10 months before the second CI than for those who stopped using their HA 3 years or more before the second CI. Especially on the WRS and SDS under the noise, there were significant differences between these two groups (P=0.01* on SDS and P=0.04* on WRS).

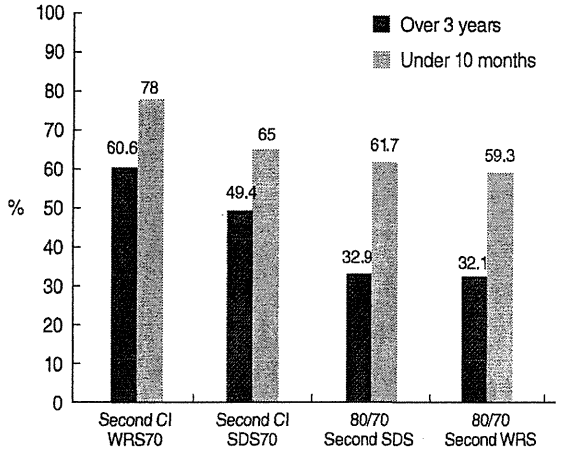


Fig. 18. Comparison of the mean word recognition score (WRS) and speech discrimination score (SDS) at 70 dB SPL at 1 m from front speaker, the mean SDS and WRS under the noise (S/N=80/70) on second cochlear implantation (CI) for those that had not used their hearing aid (HA) for over 3 years before second CI with those that had used their HA within 10 months before second CI. SPL, sound pressure level.

DISCUSSION

In all children, the WTH using the second CI was almost the same using the first CI ranging from 25 to 35 dB HL. Also, the WTH using the second CI recovered compared to the WTH using HA before their second CI (*P*=0.03*). A previous report (6) also describes that aided thresholds give better performance.

At every age, a second CI is very effective. However, the results of under 9 years old were better than the results of over 9 years old on the mean SDS under noise (S/N=80/70) on the second CI (*P*=0.04*). These results may be due to brain plasticity of the children for acquiring speech understanding under the noise (10, 14).

About use of a HA in their opposite side of first CI, on the WRS and SDS under the noise, there were significant differences between the group of over 3 years and the group of under 10 months of HA non user before second CI (*P*=0.01* on SDS and *P*=0.04* on WRS). We recommend wearing hearing aids on the opposite side after first CI. As the Japanese language uses lower frequencies, a little wearing threshold of usable frequencies remains. Also, the input from the hearing aid is very important. It is a waste to remove the HA and let the input on the opposite side of the first CI.

Most of the speech understanding scores (WRS and SDS) for children who have undergone at least 1 year habilitation after first CI and now have been fitted with a second CI show similar results to the first CI. Though the second CI eventually caught up with the first CI, it took nearly over one year.

Binaural hearing using bilateral CI is better than the first CI in all speech understanding tests. Especially, there were significant differences between the results of the first CI and bilateral CI on: 1) SDS at 70 dB SPL (*P*=0.02*); 2) SDS at 1 m from second CI side SP at 60 dB SPL (*P*=0.02*); 3) WRS at 1 m from second CI side SP at 60 dB SPL (*P*=0.02*); 4) SDS at 1 m from front SP under the noise (S/N=80/70, +10) (*P*=0.01*); 5) WRS at 1 m from front SP under the noise (S/N=80/70, +10; *P*=0.002**).

These results may show important binaural effectiveness such as binaural summation (1, 4, 5) and head shadow effect (2, 3).

Binaural summation (1, 3, 4, 7) and head shadow effect (2-5) are very likely to be important phenomena providing effective binaural advantages. Furthermore, binaural squelch (2, 4, 5) and sound localization (8, 9) are also well known to yield binaural advantages. In particular, in infancy there are many cases where the ability of hearing under the noise is very important for speech/language development.

The improvement of sound localization and hearing under noise that is provided in binaural hearing shows strong effectiveness in a typical infant environment and for children in a classroom setting (14). Bilateral CI is a very useful medical intervention for children with severe-to-profound hearing loss in Japan and elsewhere.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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Original Article

The Usefulness of Reconstructed 3D Images in Surgical Planning for Cochlear Implantation in a Malformed Ear with an Abnormal Course of the Facial Nerve

Minoru Hara, MD[1] · Haruo Takahashi, MD[1] · Yukihiko Kanda, MD[2]

[1]*Department of Otorhinolaryngology Head and Neck Surgery, Nagasaki University Hospital, Nagasaki;*
[2]*Kanda ENT Clinic, Nagasaki Bell Hearing Center, Nagasaki, Japan*

Objectives. It is not unusual for a cochlear implantation (CI) candidate to have some type of ear malformation, in particular an abnormal course of the facial nerve (FN). In this study, we attempted to reconstruct a three-dimensional (3D) image of temporal bone structures with malformation using computed tomography (CT) imaging and examined its usefulness in the surgical planning of CI in a malformed ear.

Methods. We prepared 3D images for 6 separate CI cases before surgery. First, we manually colored preoperative CT images using Photoshop CS Extended. We then converted the colored CT images to 3D images using Delta Viewer, freeware for Macintosh. Before surgery, we discussed any problems anticipated based on the 3D images and plans for surgery with those who would be performing the CI.

Results. Case 1: The subject was a 3-year-old boy with malformed ossicles, semicircular canal (SC) hypoplasia, internal auditory canal stenosis, and an abnormal course of the FN. 3D image indicated that the stapes were absent, and the FN was more anteriorly displaced, so that it was difficult to perform cochleostomy. The surgical findings were similar to those depicted on the 3D image, so we could insert an electrode based on the preoperative image simulation without complications. Case 2: The subject was a 7-year-old boy with malformed stapes, atresia of the round window, cochlear and SC aplasia, and an abnormal course of the FN with bifurcation. CI was performed with no problems, in the same manner as in Case 1.

Conclusion. We were able to successfully depict the structures of the inner ear, ossicles, and FN as 3D images, which are very easy to understand visually and intuitively. These 3D images of the malformed ear are useful in preoperative image simulation and in surgical planning for those performing a CI procedure.

Key Words. *Three-dimensional image, Temporal bone, Facial nerve anomaly, Cochlear implantation*

INTRODUCTION

A number of cochlear implantation (CI) candidates have ear malformations, in particular an abnormal course of the facial nerve (FN). The course of the FN is very important for CI surgery; the more abnormal the course of the FN, the more difficult it is to perform CI surgery. However, it is extremely difficult for surgeons to understand the three-dimensional (3D) course of the FN prior to surgery through the use of two-dimensional (2D) computed tomography (CT) images.

In this study, we attempted to reconstruct a 3D image of temporal bone structures using CT imaging for a plan of CI surgery in a malformed ear and show the 3D images of the two cases.

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• Corresponding author: **Minoru Hara, MD**
Department of Otorhinolaryngology Head and Neck Surgery, Nagasaki University Hospital, 1-7-1 Sakamoto, Nagasaki 852-8102, Japan
Tel: +81-95-819-7349, Fax: +81-95-819-7352
E-mail: harami@nagasaki-u.ac.jp

MATERIALS AND METHODS

From 2010 to 2011, we prepared 3D images for 6 separate CI cases prior to surgery. These cases consisted of 5 patients (3 boys, 2 girls, ages 1-7 years), including one bilateral case. All of the patients had congenital malformations in the temporal bone structures, such as in the FN, cochlea, semicircular canal (SC), or stapes. We created 3D images using a method we developed that utilized a public personal computer.

First, we manually colored preoperative CT images, which were axial sections of the temporal bone scanned by a normal CT scanner (SOMATOM Definition; Siemens Medical, München, Germany) at a slice thickness of 0.5 mm using Photoshop CS extended. The extended version can import and edit DICOM files directly. The inner ear labyrinth, auditory ossicles, and FN were shaded in blue, red, and yellow, respectively, maintaining the shape of these structures. We had to paint every axial section of temporal bone CT images; there were about 30-40 slices. It took about two hours to accomplish the coloring process.

We then converted the colored 2D-CT images to 3D images using Delta Viewer (DV), a freeware for Macintosh available on the Internet (http://delta.math.sci.osaka-u.ac.jp/DeltaViewer/index.html). This 3D reconstruction can be done from CT images of any condition, such as the thickness of the slice of images, but the thinner the CT slices are, the more detailed and smoother the 3D images will be. This DV-3D rendering process was completed automatically within a few minutes. In this paper, we refer to the 3D images created using DV as DV-3D images.

Fig. 1 is a DV-3D image of the normal temporal bone structures. We can rotate DV-3D images freely using the DV application.

Before each CI procedure, we discussed any problems anticipated based on the DV-3D images and planned the surgery with those who would be performing the procedure. We also brought either the printed images or the notebook PC to the operating room and compared the images with the surgical findings during the CI procedure (Fig. 2).

RESULTS

Case 1. Left ear of a three-year-old boy

The patient presented with bilaterally malformed ossicles, SC hypoplasia, internal auditory canal stenosis, and an abnormal course of the FN. We had already performed CI surgery on the

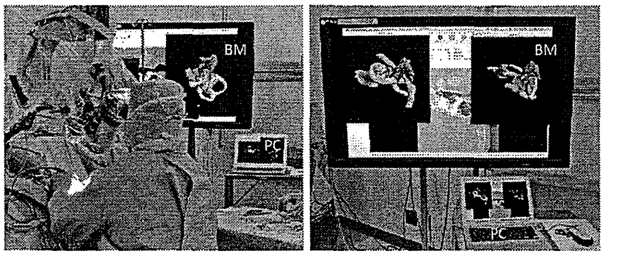

Fig. 2. The pictures of an operation room during a cochlear implantation surgery. The surgeon sees Delta Viewer 3 dimensional images displayed on the bedside monitor (BM), which is controlled by the notebook PC (Macintosh), and compares the images with surgical findings.

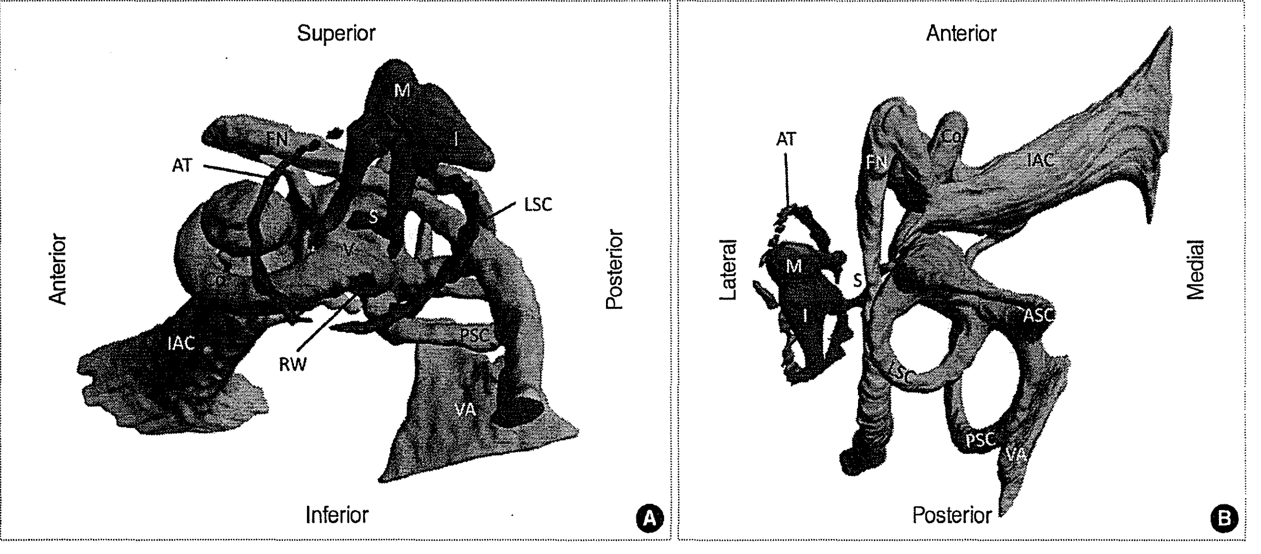

Fig. 1. Example of Delta Viewer 3 dimensional image of normal temporal bone structures of left ear based on computed tomography image. Image (A) is antero-lateral inferior view, and image (B) is superior view. The bony labyrinth was shaded in blue and includes the cochlea (Co), vestibule (V), anterior semicircular canal (ASC), lateral semicircular canal (LSC), posterior semicircular canal (PSC), and round window (RW). The internal auditory canal (IAC) and facial nerve (FN) are shaded in yellow. The ossicles are shaded in red and include the malleus (M), incus (I), and stapes (S). The annulus tympanicus (AT) and vestibular aqueduct (VA) are shaded in green and purple, respectively.

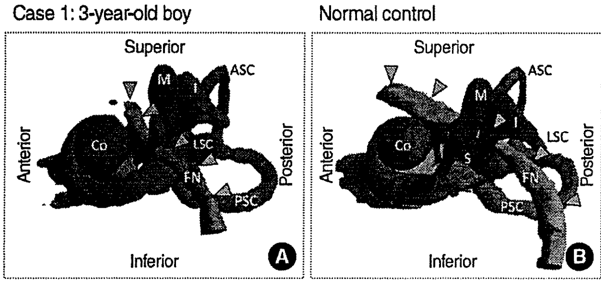

Fig. 3. (A) Lateral view of the temporal bone structures of case 1. The lateral semicircular canal (LSC) is hypoplastic, and the crus of the incus (I) and stapes (S) are absent. The labyrinthine segment of the facial nerve (FN) and the geniculate ganglion are posteriorly displaced, and the tympanic and mastoid segments of the FN are antero-inferiorly displaced, running more vertically than normal control. The cochlea (Co), malleus (M), anterior semicircular canal (ASC), and posterior semicircular canal (PSC) are intact. (B) Lateral view of normal control. The green arrowhead shows the course of the FNs.

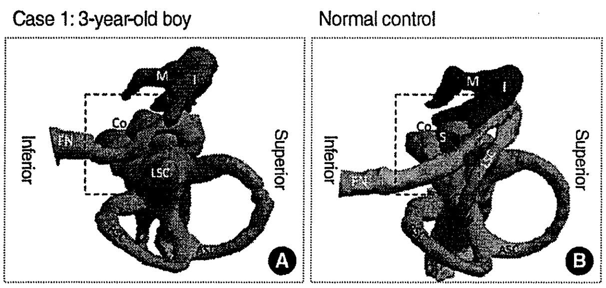

Fig. 4. (A) The Delta Viewer 3 dimensional (DV-3D) image of case 1 in the same position during cochlear implantation surgery. (B) The DV-3D image of normal control in the same position as (A). Rectangle of dashed line shows the area of Fig. 5. Co, cochlea; ASC, anterior semicircular canal; PSC, posterior semicircular canal; LSC, lateral semicircular canal; FN, facial nerve; M, malleus; I,incus; S, stapes.

patient's right ear one year before. The outcome of his speech was not as good as that of a patient with a non-malformed ear. We then planned a second CI procedure on the patient's left ear. We expected that it was going to be difficult to perform cochlear fenestration because of the facial nerve abnormality, so we prepared DV-3D images of this case before the CI surgery.

The DV-3D images (Fig. 3) indicate that the cochlear turn is intact, the lateral SC is hypoplastic, the long crus of the incus and stapes is absent, the labyrinthine segment of the FN is more posteriorly placed, and the tympanic and mastoid segments are antero-inferiorly displaced and running more vertically than normal. Fig. 4 shows the structures pictured in the same position as we found them during surgery. These images show that the stapes are absent and that the FN runs antero-inferiorly onto the oval window.

Fig. 5 shows the preoperative DV-3D image and the actual picture of the surgical findings. The space for cochlear fenestration was very narrow; however, we were able to insert an electrode based on the preoperative DV-3D image without any complications, such as FN palsy or stimulation. Fig. 6 shows an X-ray of the electrode: MED-EL, standard.

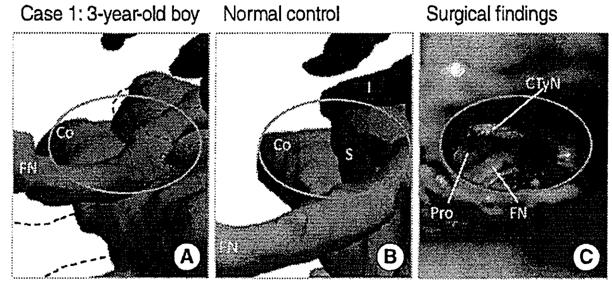

Fig. 5. The images of the surgical field of posterior tympanotomy as shown by the orange oval. (A) Case 1: close-up image of the rectangle of Fig. 4A. The stapes (S) and crus of the incus (I) are absent, and the facial nerve (FN) is antero-inferiorly displaced. Dashed line shows the position of the ossicles and facial nerve (FN) of normal control. The visible area of the basal turn of cochlea (Co) is smaller than normal. (B) Normal control: close-up image of the rectangle of Fig.4B. (C) The picture of the surgical findings of case 1. The visible area of the promontory (Pro) for cochleostomy is narrow and surrounded by the chorda tympani nerve (CTyN) and facial nerve (FN) displaced anteriorly.

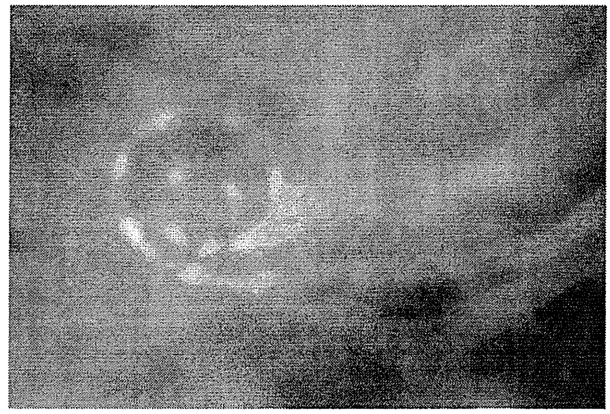

Fig. 6. The X-ray of inserted electrode of case 1.

Case 2: Left ear of a seven-year-old boy

The patient presented with bilateral cochleo-vestibular malformations and abnormalities of the stapes and course of the FN. We had already performed CI surgery on the right ear one year before. We planned a second CI procedure on this patient's left ear for the same reason we performed surgery on the first patient.

The preoperative DV-3D images (Figs. 7, 8) indicate that the shape of the cochlea and the SC are hypoplastic, the stapes seem to be fused to the FN, and the course of the FN is abnormal, as in the previous case, with bifurcation. In addition, the round window niche is closed, so there is no landmark for cochlear fenestration. We were able to successfully perform the cochlear fenestration right next to the FN, which was detected using a FN monitor and referring to a DV-3D image of the nerve course.

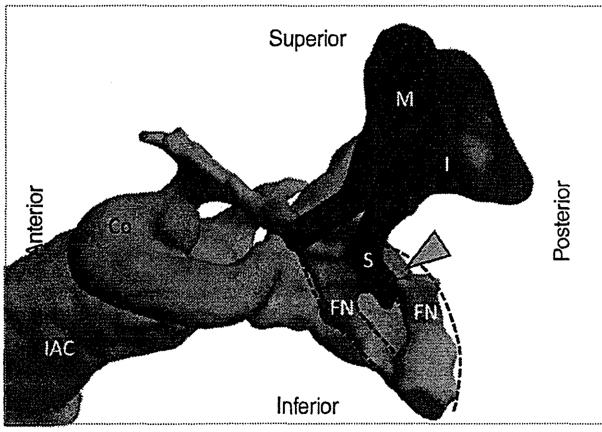

Fig. 7. Antero-lateral view of the temporal bone structures of case 2. The shape of the cochlea (Co) and semicircular canals are hypoplastic. The labyrinthine segment of the facial nerve (FN) and geniculate ganglion are posteriorly displaced, the tympanic and mastoid segments of the FN are antero-inferiorly displaced with bifurcation, and the stapes is fused to the facial nerve (arrow head). IAC, internal auditory canal; M,malleus; I, incus; S, stapes.

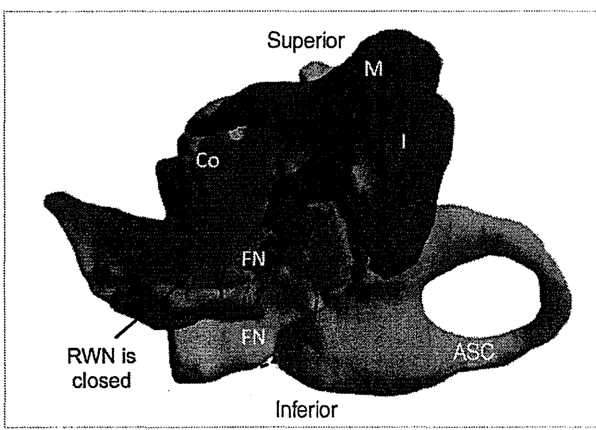

Fig. 8. The Delta Viewer 3 dimensional image of case 2 in the same position during cochlear implantation surgery. The surface of the tympanic cavity is displayed in green. The round window niche (RWN) is closed, so there was no landmark for cochlear fenestration. Co, cochlea; FN, facial nerve; ASC, anterior semicircular canal; M, malleus; I, incus.

Fig. 9 shows an X-ray of the electrode: Cochlear, standard.

Table 1 consists of a list of the DV-3D images we prepared for 6 separate CI cases that involved ear malformation; in particular, an abnormal course of the FN. In all of the cases, we were able to successfully insert electrodes by referring to DV-3D images with no technical problems. Table 1 suggests that the anomaly of the stapes indicates an abnormal course of the FN.

DISCUSSION

We prepared 3D images of patients' ear malformations prior to performing CI procedures on these patients. These 3D images contributed to the successful insertion of electrodes during surgery on these patients, each of whom had an abnormal course of the FN. There is no previous report on the clinical use of 3D imaging for preoperative planning of CI surgery.

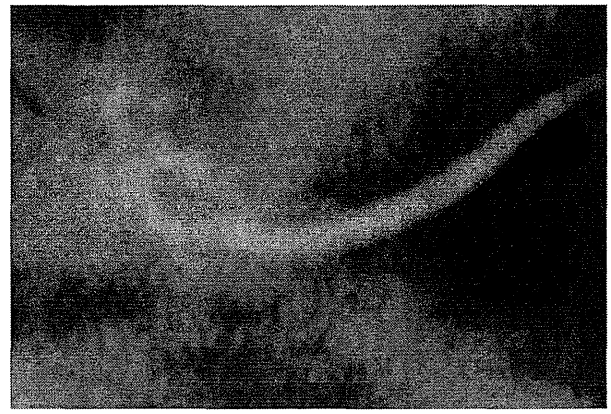

Fig. 9. The X-ray of inserted electrode of case 2.

We have been able to produce preoperative 3D images using a method we were able to develop at a low cost. Several software products are available that enable us to convert 2D medical images, such as those produced through CT or magnetic resonance imaging, to 3D. However, these imaging techniques are not suitable for creating 3D images of the temporal bone structures, because their resolution is so rough that they were developed generally for use on larger organs such as the lungs, large arteries, etc.

In very recent years, a high-performance multi-slice CT scanner and workstation have made it possible to generate detailed 3D images of the temporal bone structures for radiology diagnostics (1). Some previous articles have reported on the creation of 3D images based on a histological specimen for educational purposes (2-6).

A novelty of our method is that it enabled us to convert structures of different densities-bone structures, such as the ossicles, and soft tissue structures, such as the FN and cochlea-in the same manner on one 3D image. It is especially difficult to reconstruct a FN as a 3D image, because it is difficult to select the FN on a CT image. There is a previous report that investigated automatic FN selection through the use of special computer software; however, this method has not yet been put into use (7). There is room for further improvement in the accuracy of the DV-3D image; however, even now it is quite useful for planning a CI procedure and for avoiding surgical complications.

In this study, we suggest that an anomaly of the stapes indicates an abnormal course of the FN. It is well-established that an abnormal stapes has been associated with an anomalous course of the tympanic and mastoid segments of the facial nerve, because these are derivatives of the second branchial arch (8-10). As the labyrinthine segment of the FN is not derived from the

Table 1. Summary of cases and Delta Viewer (DV) 3D image findings

Case	Age (year) (sex)	Day of surgery (side)	DV-3D image findings					CI
			Cochlea	Semicircular canal (SC)	Stapes	Facial nerve: Labyrinthine segment	Facial nerve: Tympanic and mastoid segment	
1	3 (M)	Feb 2010 Right	Intact	Hypoplasia of LSC	Absent	Posteriorly displaced	Antero-inferiorly displaced	2nd MED-EL standard
2	7 (M)	Jul 2010 Left	Hypoplasia	Aplasia of LSC and PSC	Abnormal: fusion to FN	Posteriorly displaced	Antero-inferiorly displaced with bifurcation	2nd cochlear straight
3	5 (F)	Mar 2010 Right	Intact	Aplasia of all SCs	Absent	Posteriorly displaced	Antero-inferiorly displaced	1st MED-EL standard
4	3 (M)	Feb 2010 Right	Hypoplasia	Almost intact	Absent	Posteriorly displaced	Antero-inferiorly displaced	1st cochlear straight
5	6 (F), same patient as case 3	Feb 2011 Left	Intact	Aplasia of all SCs	Absent	Posteriorly displaced	Antero-inferiorly displaced	2nd MED-EL medium
6	1 (F)	Nov 2011 Left	Almost aplasia	Almost intact	Intact	Intact	Intact	1st MED-EL medium

CI, cochlear implantation; LSC, lateral SC; PSC, posterior SC; FN, facial nerve.

second branchial arch but from the otic cupsule, the cause of the anomaly of this segment may not be discussed on the same basis as that of the other segments related to the second branchial arch.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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Original Article

What Factors Are Associated with Good Performance in Children with Cochlear Implants? From the Outcome of Various Language Development Tests, Research on Sensory and Communicative Disorders Project in Japan: Nagasaki Experience

Yukihiko Kanda, MD[1,2] · Hidetaka Kumagami, MD[2] · Minoru Hara, MD[3] · Yuzuru Sainoo, MD[3] · Chisei Sato, MD[3]
Tomomi Yamamoto-Fukuda, MD[2] · Haruo Yoshida, MD[2] · Akiko Ito[1] · Chiharu Tanaka[1] · Kyoko Baba[1]
Ayaka Nakata[1] · Hideo Tanaka[1] · Kunihiro Fukushima, MD[4] · Norio Kasai, MD[4,5] · Haruo Takahashi, MD[2]

[1]Kanda ENT Clinic, Nagasaki Bell Hearing Center, Nagasaki; [2]Department of Otolaryngology Head and Neck Surgery, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki; [3]Department of Otolaryngology Head and Neck Surgery, Nagasaki University Hospital, Nagasaki; [4]Department of Otolaryngology Head and Neck Surgery, Okayama University Postgraduate School of Medicine, Dentistry, and Pharmaceutical Science, Okayama; [5]The Association for Technical Aids, Tokyo, Japan

Objectives. We conducted multi-directional language development tests as a part of the Research on Sensory and Communicative Disorders (RSVD) in Japan. This report discusses findings as well as factors that led to better results in children with severe-profound hearing loss.

Methods. We evaluated multiple language development tests in 33 Japanese children with cochlear implants (32 patients) and hearing aid (1 patient), including 1) Test for question and answer interaction development, 2) Word fluency test, 3) Japanese version of the Peabody picture vocabulary test-revised, 4) The standardized comprehension test of abstract words, 5) The screening test of reading and writing for Japanese primary school children, 6) The syntactic processing test of aphasia, 7) Criterion-referenced testing (CRT) for Japanese language and mathematics, 8) Pervasive development disorders ASJ rating scales, and 9) Raven's colored progressive matrices. Furthermore, we investigated the factors believed to account for the better performances in these tests. The first group, group A, consisted of 14 children with higher scores in all tests than the national average for children with hearing difficulty. The second group, group B, included 19 children that scored below the national average in any of the tests.

Results. Overall, the results show that 76.2% of the scores obtained by the children in these tests exceeded the national average scores of children with hearing difficulty. The children who finished above average on all tests had undergone a longer period of regular habilitation in our rehabilitation center, had their implants earlier in life, were exposed to more auditory verbal/oral communication in their education at affiliated institutions, and were more likely to have been integrated in a regular kindergarten before moving on to elementary school.

Conclusion. In this study, we suggest that taking the above four factors into consideration will have an affect on the language development of children with severe-profound hearing loss.

Key Words. *Cochlear implant, Children, Research on sensory and communicative disorders, Language development, Japan*

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• Corresponding author: **Yukihiko Kanda, MD**
Kanda ENT Clinic, Nagasaki Bell Hearing Center, Wakakusa 4-25, Nagasaki 852-8023, Japan
Tel: +81-95-841-7038, Fax: +81-95-841-7041
E-mail: n-bell@estate.ocn.ne.jp

INTRODUCTION

Cochlear implantation (CI) is a highly specialized medical procedure for severe-to-profound hearing loss in patients all over the world. Newborn hearing screening (NHS) makes early detection and thus early intervention possible. NHS has allowed us to test 95% of newborns in Nagasaki over the last 4 years. With the rapid increase in use of pediatric CI, there is a need to develop more intensive, longitudinal, and standardized tests for auditory, speech, and communication skills and language development. There are very few packages that include multiple language development in the world.

As a part of the Research on Sensory and Communicative Disorders (RSCD) project in Japan, we examined various language development tests for children fitted with cochlear implants. This report discusses findings as well as factors that led to better results in children with severe-profound hearing loss.

METHODS AND RESULTS

Subjects

We examined 33 Japanese children (32 cochlear-implant patients and 1 hearing-aid patient) in our hearing center for the RSCD project. Children were selected according to the following criteria: 1) aged between 48 to 155 months and 2) congenital hearing impairment with a hearing level >70 dB (average over multiple frequency bands). Children unable to complete these tests because of further disabilities were not included. A consent form was provided in 2009. The age distribution was as follows: 4 years of age (4); 5 years (5); 6 years, i.e., 1st grade in primary school (4); 7-8 years, 2nd grade (2); 8-9 years, 3rd grade (4); 9-10 years, 4th grade (6); 10-11 years, 5th grade (4); and 11-12 years, 6th grade (4). Only one patient used hearing aids in both ears, and the remaining 32 children wore cochlear implants. Ten children had gone through the NHS process, while the other 23 had not. The age at fitting of hearing aids varied from 4 months to 5 years 4 months, and the age of cochlear implant surgery varied from 1 year 6 months to 6 years 3 months.

The tests were conducted between April 2009 and March 2010.

Methods 1

We asked the children to perform the following tests:

- Test for question and answer interaction development (TQAID): This test aims to evaluate interpersonal communication skills (IPCS) with 57 questions divided into 10 categories.
- Word fluency test (WFT): This test was conducted as a productive vocabulary task. Children were asked to generate as many words as possible from a given category in 60 seconds.
- Japanese version of the Peabody picture vocabulary test-revised (PVTR).
- The standardized comprehension test of abstract words (SCTAW): This test was conducted as comprehensive vocabulary tasks, and these consist of 32 or 45 abstract words selected from Japanese textbooks.
- The screening test of reading and writing for Japanese primary school children (STRAW): This test was also conducted to examine the children's reading and writing abilities. Since preschool children have not yet learned Katakana or Kanji characters, the test for these children only included Hiragana characters.
- The syntactic processing test of aphasia (STA): The STA, a syntax test that is like the test for the reception of grammar (TROG) for Japanese language users, is a test that evaluates the comprehension and production ability of syntactic structures.
- Criterion-referenced testing (CRT) for Japanese language and mathematics.
- Pervasive development disorders ASJ rating scales (PARS) to determine autistic tendency.
- Raven's colored progressive matrices (RCPM).

Results 1

The results showed that children suffering from hearing loss exceeded the national average of all children with hearing difficulties by at least 60.6% and up to 100% (Fig. 1). A total of 76.2% of all scores exceeded the national average of children with hearing difficulties. On the CRT for Japanese language and mathematics, 70.0% of all scores exceeded the national average of scores obtained by normal-hearing children (Fig. 2). We investigated the factors believed to account for the better performances in these tests.

Methods 2

To determine the factors that allowed the children reported un-

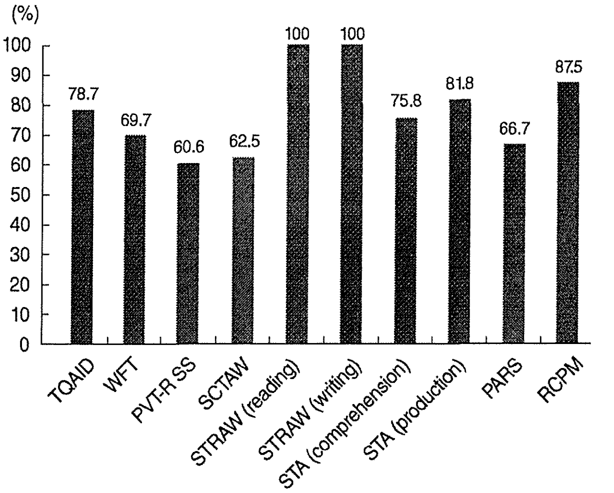

Fig. 1. The results of the various language development tests. The results show that children suffering from hearing loss exceeded the national average of all children with hearing difficulties by at least 60.6% and up to 100%. TQAID, test for question and answer interaction development; WFT, word fluency test; PVTR, Peabody picture vocabulary test-revised; SCTAW, standardized comprehension test of abstract words; STA, syntactic processing test of aphasia; PARS, pervasive development disorders ASJ rating scales; RCPM, Raven's colored progressive matrices.

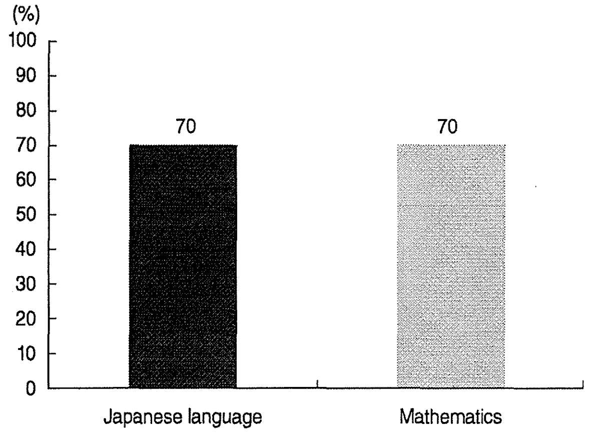

Fig. 2. On the criterion-referenced testing for Japanese language and mathematics, 70.0% of all scores exceeded the national average of scores obtained by normal-hearing children.

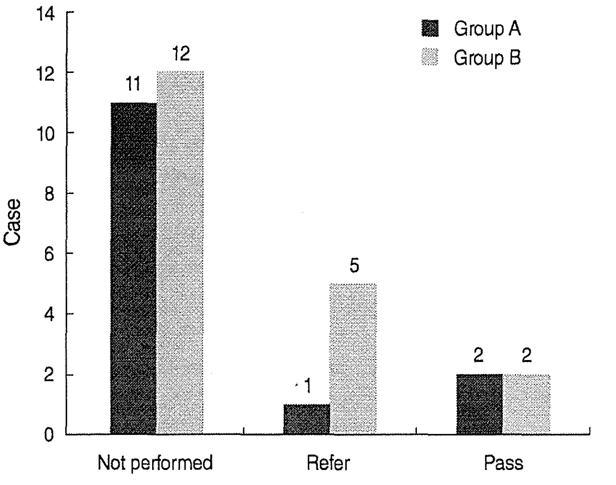

Fig. 3. Whether or not the child went through newborn hearing screening.

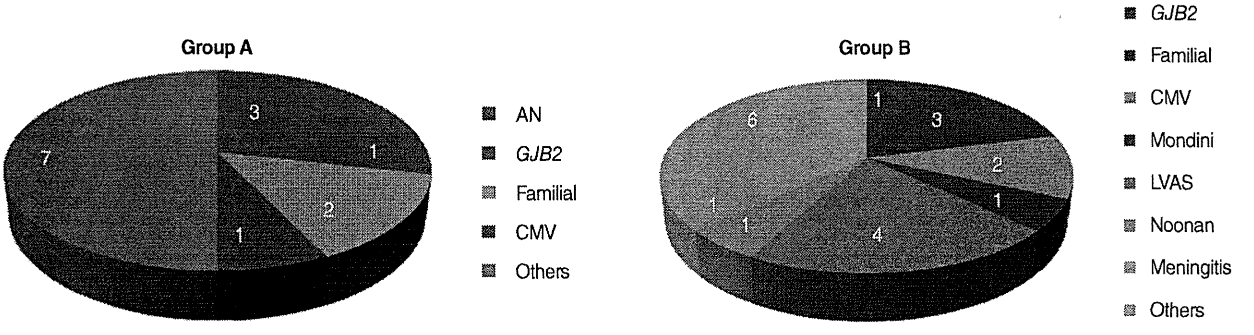

Fig. 4. Causes of deafness. AN, auditory neuropathy; *GJB2*, Gap junction protein, beta-2, 26kDa (*GJB2*) gene mutation; CMV, congenital cytomegalovirus infection; LVAS, large vestibular aqueduct syndrome.

der Results 1 to have better results, we divided the children into two groups. The first group, group A, consisted of 14 children with higher scores in all tests than the national average for children with hearing difficulty. The second group, group B, included 19 children that scored below the national average in any of the tests.

Determining criteria within each group were as follows: 1) whether the child had gone through NHS, 2) the cause for the hearing loss, 3) the age at which the child began to wear hearing aids, 4) the age at which the child received CI, 5) number of visits to our hearing center since initial examination, 6) the amount of time since CI, 7) current average hearing level, 8) current average wearing threshold, 9) whether the child has any siblings, 10) amount of time spent studying at home on a daily basis, 11) educational method (school), 12) the period of integration and the period of auditory verbal/oral education, 13) educational institution child attended before entering primary school.

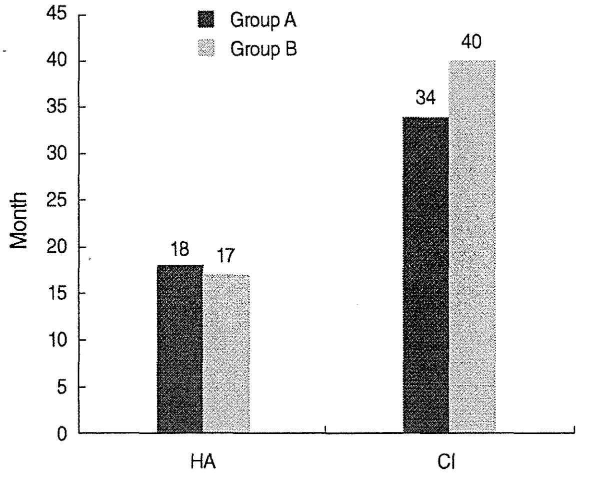

Fig. 5. The mean age for children to start wearing a hearing aid (HA) and cochlear implant (CI).

Results 2

Quite a few children underwent NHS (Fig. 3). Causes for deafness are shown in Fig. 4. There were many cases of inner ear and cochlear nerve anomaly and developmental disabilities in group B. There were no significant differences between the two groups in terms of the mean age for children to start wearing a hearing aid or the mean age for CI (Fig. 5).

The mean period of the visit at our hearing center was significantly longer in group A than in group B (P=0.049 <0.05*) (Fig. 6). The mean wearing period for the cochlear implant was significantly longer in group A than in group B (P=0.02*) (Fig. 6). The mean of the current average hearing level on their CI side was 115 dBHL for group A and 113 dBHL for group B on their CI side. On the non-operation side, it was 102.1 dBHL for group A and 97.1 dBHL for group B. The mean of the present average wearing threshold was 26.8 dBHL for group A and 28.2 dBHL for group B on their CI side. On the non-operation side, it was 59.2 dBHL for group A and 59.3 dBHL for group B. There were no significant differences in these results between the two groups (Fig. 7). Children in group A were more likely to have older siblings; however, there was no significant difference between groups

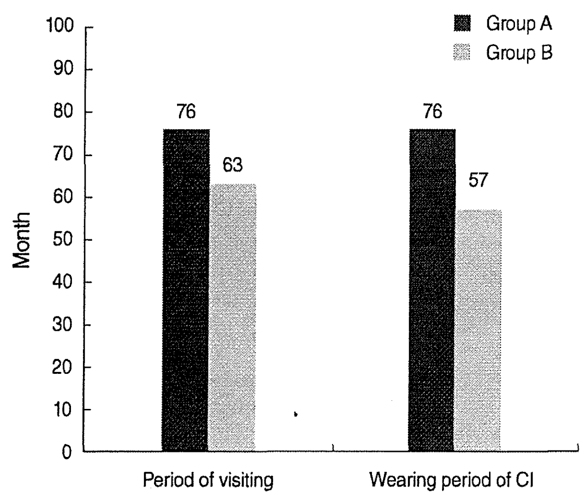

Fig. 6. The mean period of the visit at our hearing center and the mean wearing period of cochlear implant (CI).

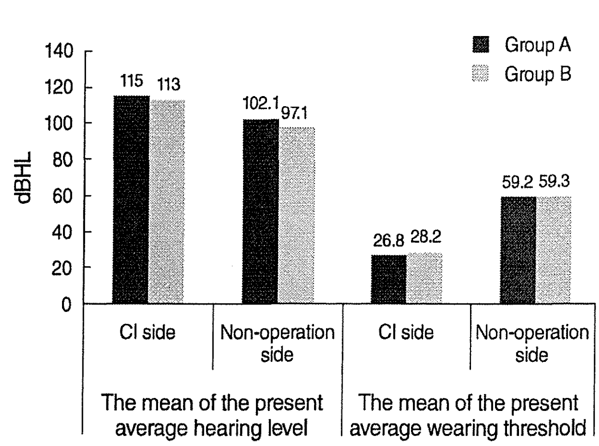

Fig. 7. The mean of the current average hearing level and the present average wearing threshold.

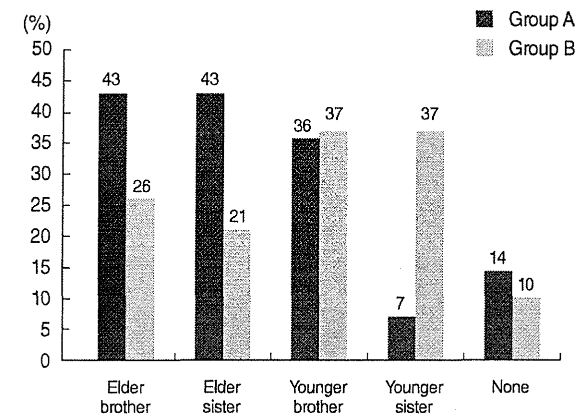

Fig. 8. Whether the child has any siblings.

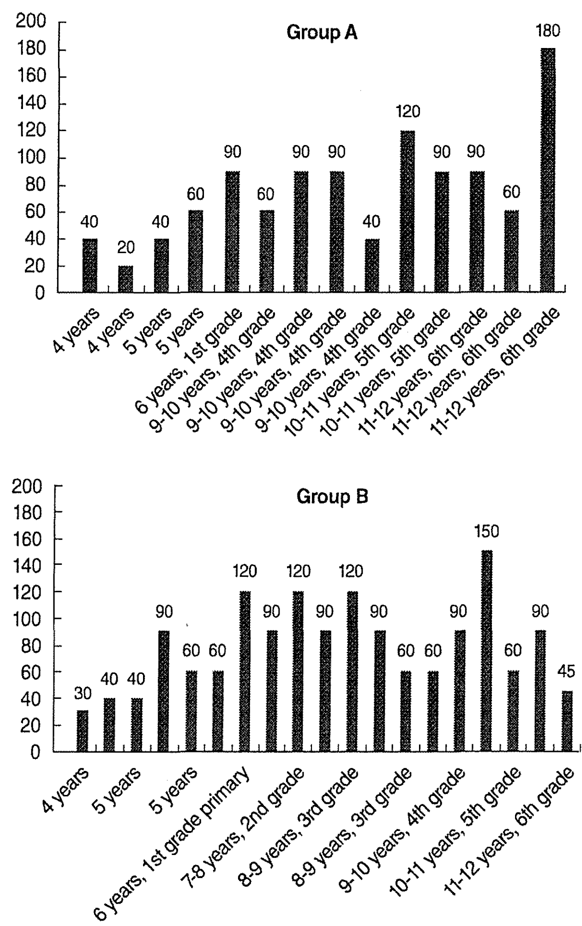

Fig. 9. The amount of time spent studying at home on a daily basis.

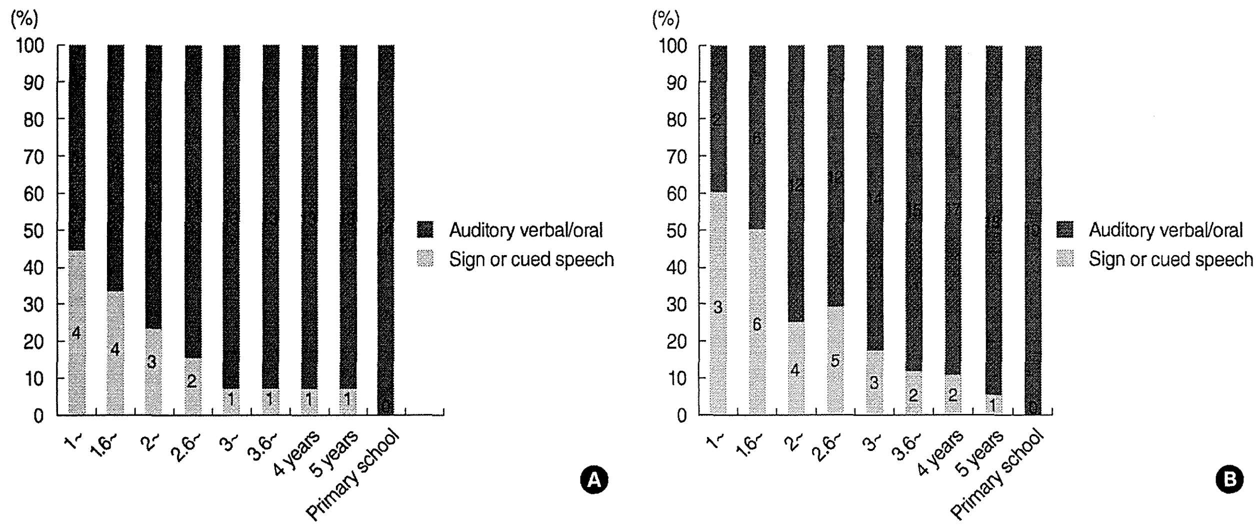

Fig. 10. Educational method (school). A, group A; B, group B.

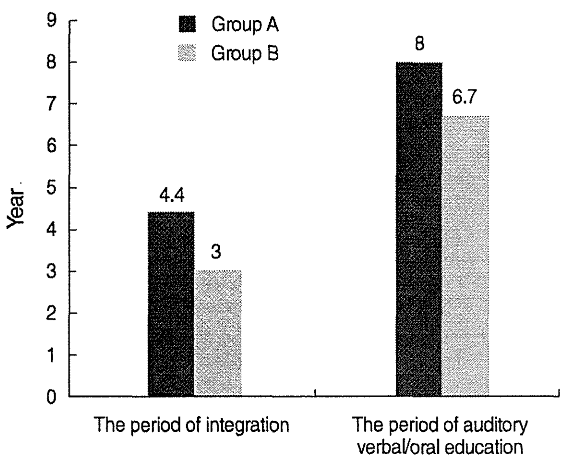

Fig. 11. The period of integration and the period of auditory verbal/oral education.

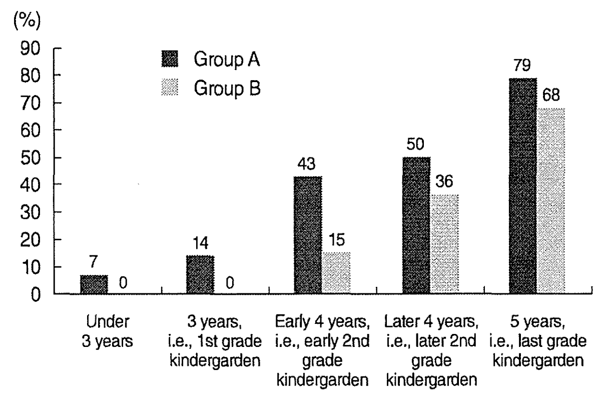

Fig. 12. Educational institution child attended before entrance to primary school.

A and B (Fig. 8). The mean amount of time spent studying at home on a daily basis was 76.4 minutes for group A and 79.2 minutes for group B; these times were not significantly different (Fig. 9). From the age of 1 year to the end of preschool, the education for group A concentrated on auditory verbal and/or oral methods, while that for group B was geared towards sign or cued speech type education; there were significant differences between groups A and B (P=0.003 <0.01**) (Fig. 10).

Children in group A attended regular school for 4.4 years, and those in group B attended for 3 years. Auditory verbal/oral education was 8 years for group A and 6.7 years for group B. While group A's education was longer than that of group B, there were no significant differences between the two groups (Fig. 11). Fig. 12 shows the percentage of children who were integrated into regular kindergarten and nursery school before attending elementary school; there were significant differences between groups A and B (P=0.01*).

DISCUSSION

What factors are associated with good performance in language development in children with cochlear implants? It is very important to gauge the effectiveness of the appropriate intervention for hard-of-hearing infants. Research and evaluation of language development for children with cochlear implants have been conducted and should continue. However, there are very few packages that include multiple language development in the world (1-8).

In 2010, Fukushima et al. planned to assess the current status of hearing impaired children in Japan using the RSCD project, and many tests were used as a part of this nationwide research