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Surgical Outcomes of Simultaneous Cochlear Implantation and Intracochlear Schwannoma Removal

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Abstract

Objective. Intracochlear schwannoma is very rare, and complete loss of hearing is inevitable after the removal of this tumor. Here, we discuss cochlear implantation (CI) performed simultaneously with the removal of an intracochlear schwannoma.

Study Design. Retrospective single-center study.

Setting. Tertiary medical institute.

Methods. Simultaneous CI and intracochlear schwannoma removal were performed in 4 subjects. After subtotal cochleostomy, the tumors were removed meticulously, with preservation of the modiolus. A new slim modiolar electrode (Nucleus CI632) was placed in a manner that hugged the modiolus. The surgical outcomes of functional gain, word recognition score (WRS), sound localization, and hearing in noise and speech intelligibility tests were investigated.

Results. Intracochlear schwannomas were removed successfully from the 4 patients, with no remnant tumor. The mean aided hearing threshold 6 months after surgery was 25.0 ± 1.8 dB, and the mean-aided WRS with a 60 dB stimulus was 36.0 ± 18.8% (range 16%-60%). The Categorical Auditory Performance (CAP) score of the 3 single-sided deafness patients under contralateral ear masking was 7. The CAP score of the patient with bilateral sensorineural hearing loss was 6, which improved from a preoperative score of 0.

Conclusion. When an intracochlear schwannoma does not completely invade the modiolus, CI with simultaneous tumor removal can be performed successfully, resulting in good hearing performance. A slim modiolar electrode can be placed stably at the modiolus after schwannoma removal.

Keywords

acoustic neuroma, cochlear implantation, intracochlear schwannoma, intralabyrinthine schwannoma, vestibular schwannoma

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Vestibular schwannoma (VS) is a slow-growing benign tumor originating from the eighth nerve and occurring mainly in the internal auditory canal (IAC) and cerebellopontine angle.[1] The incidence of VS in the United States is 1.09 per 100,000 population.[1] The treatment modalities applied to VS include close observation with serial magnetic resonance imaging (MRI) under a “wait-and-see” approach, surgical tumor resection, or stereotactic radiation therapy, depending on the tumor location and symptoms.[2-4]

When tumors originate from the peripheral branches of the cochleovestibular nerve and involve labyrinthine structures, such as the cochlea, vestibule, and semicircular canals, they are classified as intralabyrinthine schwannomas (ILS).[5] The incidence of ILS over the past decade was 0.81 per 100,000 person-years.[6] Kennedy et al and Van Abel et al presented a classification of ILS based on the location and reported that accompanying symptoms included unilateral hearing loss (99%-100%), dizziness (29%), and tinnitus (61%), often in conjunction with severe to profound hearing loss.[3,4] This severe hearing loss is of the intracochlear schwannoma type.[7]

In patients with severe to profound hearing loss, the most effective method for hearing rehabilitation is cochlear implantation (CI).[8-11] However, in patients

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with ILS and single-sided deafness (SSD), there are concerns regarding CI. Follow-up of the residual tumor requires repeated MRI and CI is problematic because it appears as an artifact in the images. Advances in CI devices could solve this issue.[12] Thus, there have been attempts to perform CI by penetrating the intracochlear schwannoma without removing it.[7,13] Simultaneous removal of an intracochlear schwannoma and CI is challenging because the tumor can invade the modiolus, which may be damaged during surgery. Tumor removal inevitably destroys the bony labyrinth, which makes electrode placement difficult. Nevertheless, it is better to perform intracochlear schwannoma resection and CI simultaneously. After removing the tumor, however, the surgeon needs to consider how to mount the electrode in the cochlea stably when a substantial portion of the lateral wall is destroyed. In this situation, the electrode should enclose the modiolus of the cochlea. To achieve this, Plotke et al used a customized CI electrode after intracochlear schwannoma removal.[14] However, we believe that the newly introduced Nucleus® CI632 slim modiolar electrode is ideal for achieving proximity to the modiolus, without the need for the customization done by Plontke et al. If the modiolar structure can be preserved, this modiolus-hugging electrode can be placed at the correct location. In this manner, we attempted simultaneous intracochlear schwannoma removal and CI. This study presents a procedure for performing the surgeries simultaneously and analyzes the audiological outcomes.

Methods

Simultaneous CI and intracochlear schwannoma removal was performed in 4 patients between December 2020 and November 2021 at the Department of Otolaryngology, Ajou University Hospital, Suwon, Republic of Korea. This study was approved by the Institutional Review Board of Ajou University Hospital (approval no. AJIRB-MED-EXP-22-286). As it was a retrospective study, the review board waived the requirement for informed consent.

Pure-tone audiometry at frequencies of 0.5 to 8 kHz and the word recognition score (WRS) for monosyllabic stimuli at 60 dB with complete masking of the contralateral ear were performed preoperatively, and at 3 and 6 months after surgery. The Hearing in Noise Test (HINT) was performed to investigate hearing function in the context of noise. In the quiet condition of the HINT, the minimum intensity required to recognize sentences (the reception threshold for speech) was measured. In the noisy environment, the signal-to-noise ratio was calculated; in the presence of noise of 65 dB(A) intensity, the threshold of sound intensity at which subjects could accurately repeat 50% of a sentence was determined along with the signal-to-noise ratio, expressed in dB(S/N), which equates to the threshold of this intensity minus 65 dB(A) (the noise intensity level). A sound localization test was administered, and the percent correct (PCT) was calculated as the rate of correct answers for each stimulus source; the mean absolute error (average angle difference between the actual sound-emitting speaker and the speaker indicated by the patient) and root mean square (RMS) was calculated. Measurements in the unaided condition were performed preoperatively.

Speech intelligibility was assessed with monosyllabic words, disyllabic words, and sentences in the sound field 3 and 6 months after surgery. During the test, the contralateral ear was masked only with an earplug.

Schwannoma involvement was evaluated using an MRI of the temporal bone. The tumor of patient #1 involved the entire cochlear structure, from the base to the apex. And that of patient #2 involved the basal to the middle turn. The tumor of patient #3 involved only the middle turn, and that of patient #4 involved only the basal turn (**Figure 1**).

Surgical Procedures

The beginning of the surgical procedure was the same as that performed for CI using the transmastoid approach; mastoidectomy and posterior tympanotomy were performed. Then, the tympanic membrane was elevated using the transmeatal approach to secure an adequate surgical view for subtotal cochleostomy. The malleus and incus were removed. The location of the round window (RW) was identified before subtotal cochleostomy.

Similar to the effect of the transmeatal approach, identification of the cochlea through the external auditory canal (EAC) could provide a better surgical view. A subtotal cochleostomy was performed from the main location of tumor involvement. The schwannoma was exposed by drilling and removal of the bony portion of the cochlea (**Figure 2A-D**). The size of the opening for the subtotal cochleostomy differed depending on the tumor size and location. The tumor was removed meticulously, with preservation of the modiolus. Because the tumor did not invade the modiolus, it could be dissected from the modiolus easily (**Figure 2A′-D′**).

All 4 patients underwent CI using the Nucleus® CI632 (Cochlear) via the conventional RW approach after tumor removal. This electrode was designed to wrap around the modiolus optimally when inserted using this approach; accordingly, it was pushed through the RW and confirmed microscopically to adhere well to the modiolus (**Figure 2A″-D″**). Intraoperative neural response telemetry was performed immediately after electrode array insertion. Patient #1 showed responses in 4 of 22 electrodes, patients #2 and #3 were reactive except for 1 electrode, and patient #4 showed full responses at all electrodes. All of the electrodes in patient #1 functioned well during the mapping procedure.

The defect in the cochlea was filled using cartilage chips and soft tissue to hold the electrode in position. Then, it was closed using cartilage with the perichondrium attached, and covered with bone pate and fascia (**Figure 3**). For the small opening in patient #3, no soft tissue or cartilage insertion

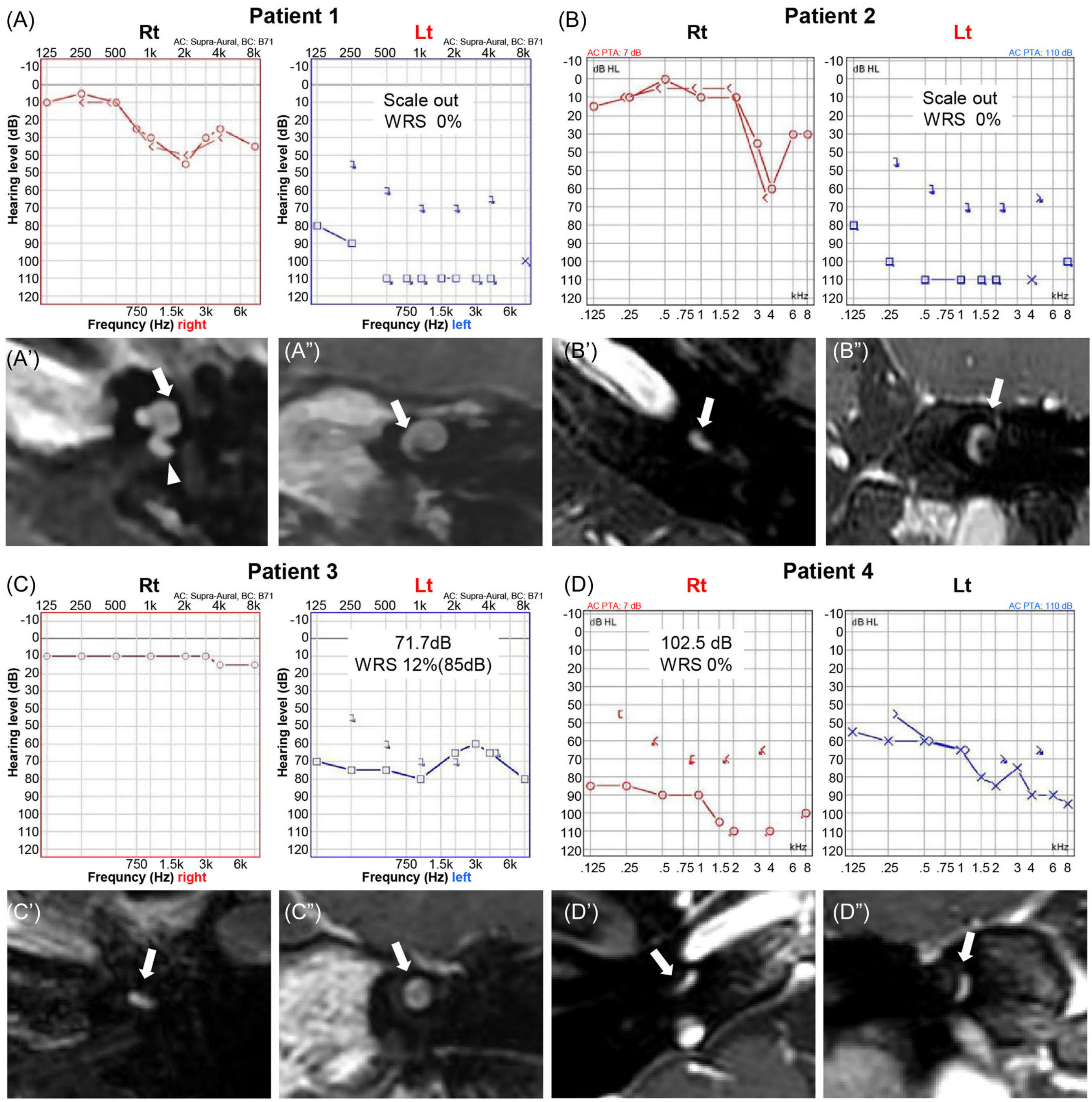

Figure 1. (A-D) Preoperative pure-tone audiometry (PTA) results and WRS. Axial (A′-D′) and coronal (A″-D″) contrast-enhanced T1-weighted MRI images. The white arrowhead in (A′) indicates vestibular involvement. HL, hearing loss; Lt, left; MRI, magnetic resonance imaging; Rt, right; WRS, word recognition scores.

into the cochlea was necessary. After the completion of all surgical procedures, the electrode position was confirmed using an intraoperative transocular view.

Results

The mean age of the 4 patients was 55.0 ± 15.1 (range 36-73) years. Three patients were male and 1 was female. Patients #1 to #3 had SSD in the left ear, in which CI was performed. Patient #4 had bilateral sensorineural hearing loss (SNHL), and the device was implanted on the right side (**Table 1** and **Figure 1**).

Patients #1, #3, and #4 had no complications after CI. Patient #2 experienced postoperative benign horizontal-canal paroxysmal positional vertigo, which was resolved by repeated canalith repositioning procedures.

Functional gain tests performed 6 months postoperatively showed that the mean hearing threshold under the

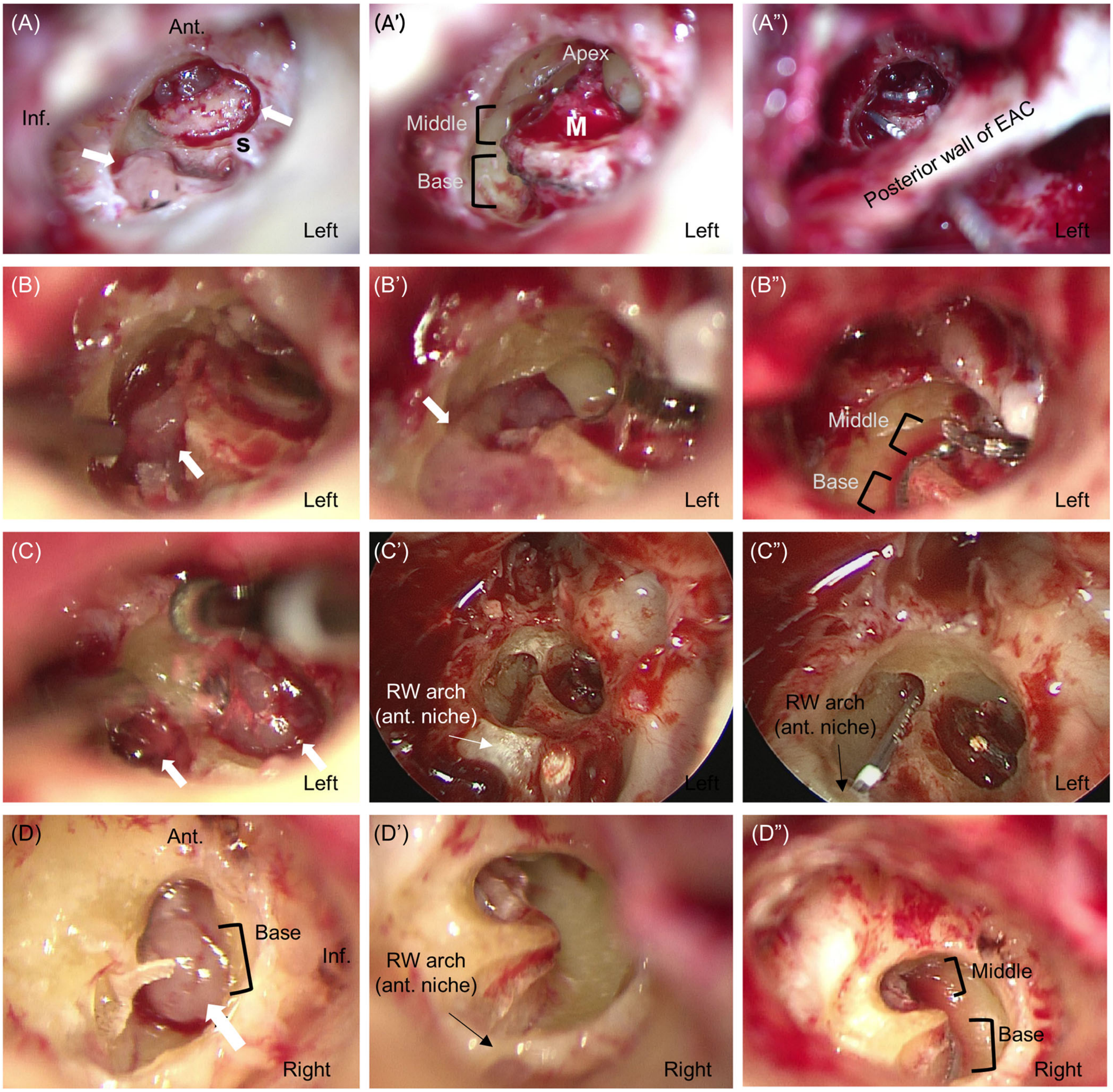

Figure 2. Intraoperative images. (A-A″) Patient #1; (B-B″) patient #2; (C-C″) patient #3; and (D-D″) patient #4. The white arrows indicate schwannomas. Ant., anterior; EAC, external auditory canal; Inf., inferior; M, modiolus; RW, round window; S, stapes head.

aided condition was 25.0 ± 1.8 dB (**Figure 4A**). The postoperative WRS at 60 dB with complete masking of the contralateral ear increased to 36.0 ± 18.8% (Figure 4B). The Categorical Auditory Performance (CAP) score was calculated and speech intelligibility (with monosyllabic words) was evaluated in SSD patients #1 to #3 at 6 months. The mean CAP score was 7 and the mean sound field monosyllable word score was 92.6 ± 3.2%. The CAP score of patient #4 (who had bilateral SNHL) increased to 6 and the sound field monosyllabic score was 64.8% at 6 months, indicating that CI effectively rehabilitated this patient's hearing (Supplemental Figure S1, available online).

For SSD patients #1 to #3, whether hearing in a noisy environment and sound localization could be improved was more important than audiological outcomes. The HINT was administered to these patients. For patient #3, the signal-to-noise ratio decreased under all but the ipsilateral noise condition (under which it increased). The other patients showed improvement under all 4 conditions (frontal, ipsilateral, contralateral, and composite noise; **Figure 4C-E**) The RMS and PCT values of patients #2 and #3 had improved at 6 months after surgery, whereas those of patient #1 had worsened. Patients #1 to #3 showed

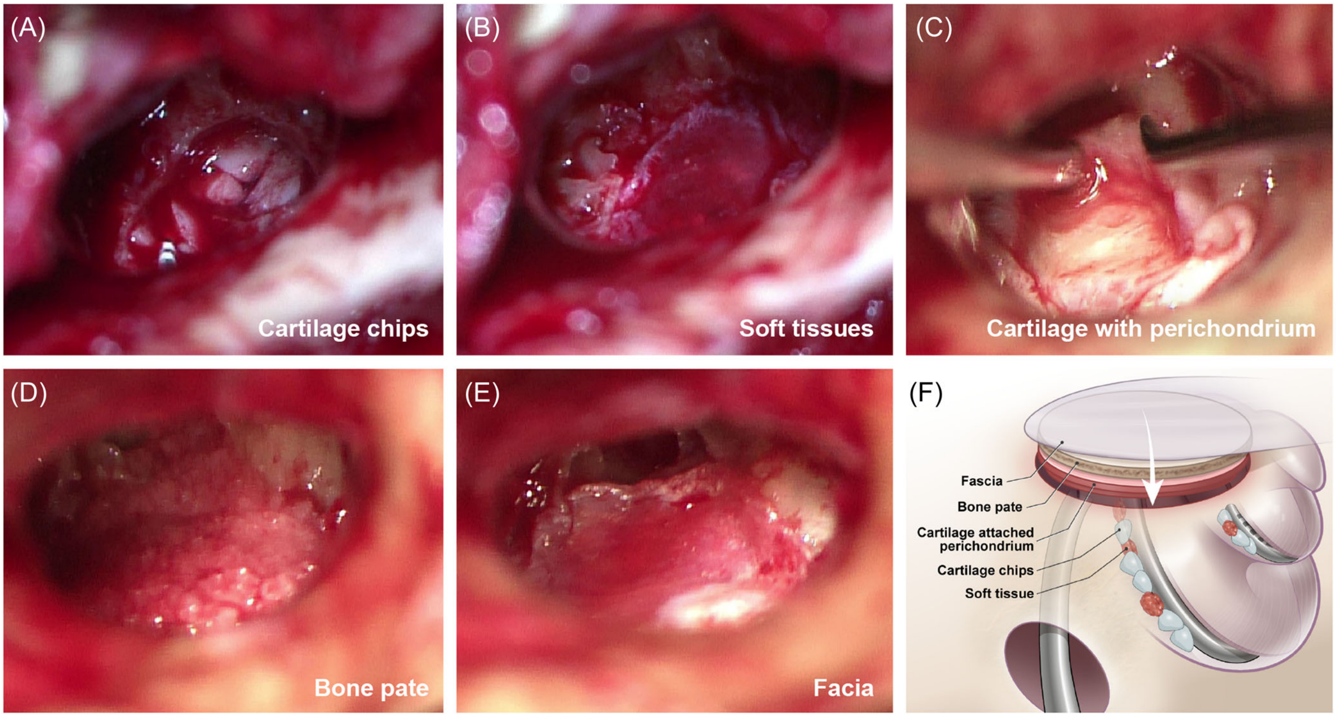

Figure 3. Step-by-step procedure for cochlear reconstruction. Cartilage chips (A), soft tissue (B), cartilage with perichondrium (C), bone pate (D), and fascia (E) were used to close the cochleostomy defects. (F) Schematic diagram of the reconstruction.

functional recovery and improvement at 1 year postoperatively (**Figure 4F**).

Discussion

The reported incidence of schwannoma in the inner ear and IAC has increased as MRI accessibility has improved.[15] As this tumor is benign, management is determined by its size and the associated symptoms. For small asymptomatic tumors, clinical observation with serial MRI is recommended. For tumors that have invaded critical areas, such as the cerebellopontine angle, gamma knife surgery, or surgical resection should be considered.[3,16,17] Surgical management may also be an option for ILS that involve the labyrinth, that is, the cochlea, vestibule, or semicircular canals. Symptoms include hearing loss, tinnitus, and vertigo. However, removal of the schwannoma does not mean that the symptoms will resolve;[17] in fact, it leads to more severe hearing loss. In one report, the hearing was preserved after removing the ILS, but the tumor in this exceptional case only involved the lateral semicircular canal.[18] In another report, the hearing was preserved after the removal of a very small tumor limited to the scala tympani near the window through an extended RW approach.[19] Since it is difficult to preserve hearing in cochlear schwannomas that invade the cochlea, it is necessary to consider how to manage this unilateral hearing loss.

Hearing rehabilitation modalities that can be chosen for SSD patients are contralateral routing of signals (CROS) hearing aids, bone conduction implants, and CI. Patients with SSD have difficulty localizing sound and understanding speech in noisy environments, as these skills are related to binaural hearing. Thus, the use of CROS hearing aids or bone conduction implants cannot be expected to improve the hearing performance of SSD patients, and CROS hearing aids confer no significant benefits.[10] By contrast, because CI can achieve binaural hearing, it is more appropriate for the auditory rehabilitation of SSD patients, including those with VSs.[20] Indeed, CI has been attempted in cases with VS.

Some reports describe the performance of CI without the removal of a VS involving the IAC. CI performance is favorable, but device artifacts may impair the visualization of the IAC and inner ear, preventing the tracking of tumor growth.[9,11,21] In a case similar to that described here, CI was attempted without intracochlear schwannoma removal; the patient's open-set speech perception was good after electrode insertion, but the authors reported that they encountered resistance during insertion due to the remaining tumor in some patients.[7] Simultaneous and staged operations for VS removal and CI are of concern. Delayed CI was attempted in a staged approach that required the additional surgical technique of dummy electrode insertion to prevent fibrosis at the tumor removal site. However, the postoperative audiological outcomes did not differ from those achieved with simultaneous surgery.[22,23] Thus, simultaneous surgery seems more appropriate.

Plontke et al and Aschendorff et al reported simultaneous CI and intracochlear schwannoma removal.[14,24,25]

Table 1. Patient Demographics

Patient no.	Age at implant (y)	Sex	Affected side	Duration of hearing loss (y)	Preoperative symptoms	Tumor location	Device	Postoperative symptoms
#1	55	F	Lt (SSD)	10	Hearing loss, dizziness, tinnitus	Whole cochlea (basal to apical turn)	CI632	-
#2	36	M	Lt (SSD)	8	Hearing loss, tinnitus	Intracochlear (basal to middle turn)	CI632	Dizziness (HC-BPPV)
#3	56	M	Lt (SSD)	3	Hearing loss, dizziness, ear fullness	Intracochlear (middle turn)	CI632	-
#4	73	M	Rt (bilateral SNHL)	2	Hearing loss, dizziness	Intracochlear (basal turn)	CI632	-

Abbreviations: F, female; HC-BPPV, horizontal canal benign paroxysmal positional vertigo; Lt, left; M, male; Rt, right; SNHL, sensorineural hearing loss; SSD, single-sided deafness.

We also performed such a procedure in the present case, in the order of subtotal cochleostomy, schwannoma removal with preservation of the modiolus, electrode array insertion, and cochlear reconstruction. Subtotal cochleostomy was performed first to expose the entire tumor, regardless of its location. Then, *en-bloc* removal of the tumor was performed using the "push-through" and "pull-through" techniques described by Plontke et al, which minimized surgical trauma to the modiolus.[24] The tumor should be removed from the apex of the cochlea in the direction of the base because the larger diameter of the basal turn may render tumor removal in the opposite direction difficult.

The most substantial difference between previously reported surgical techniques and ours may be in the insertion of the electrode array. In a recently reported case series, a custom-made device that was malleable and could be shaped manually into a spiral modiolus-hugging structure was used.[14] As this device was shaped into the form of the modiolus before "insertion," the procedure may be characterized more correctly as electrode placement over the modiolus, rather than electrode insertion into the cochlea. Such placement does not require the maintenance of the RW arch.[14] During our surgical procedure, the use of the new slim modiolar electrode enabled the maintenance of the RW structure. The electrode array was initially straight and encased in a sheath. Then, it was positioned at the RW, followed by the insertion of the modiolus-hugging electrodes. The RW structure should be maintained when inserting the new modiolar electrode, and caution should be exercised when performing the cochleostomy. For simultaneous CI and schwannoma removal surgery performed with this new slim modiolar electrode (Nucleus® CI632), the maintenance of the RW structure is better (Supplemental Figure S2, available online).

The cochlea was reconstructed to prevent electrode dislocation and increase modiolar proximity. This procedure was accomplished using multilayered reinforcement with cartilage chips, fascia, soft tissue, and surgical glue. Postoperative electrode dislocation was not suspected in any of the 4 patients studied. However, long-term follow-up is required to confirm this outcome.

In previous studies, the posterior wall of the EAC was lowered to improve surgical visibility.[26-28] However, removing the EAC made it difficult to hold the electrode array, so subtotal petrosectomy should be combined with the closure of the EAC. It would be more appropriate to perform this surgical procedure if the posterior wall of the EAC interferes with the visualization of the schwannoma. Lowering the EAC was not considered in our cases. We obtained a sufficient surgical view for both tumor removal and CI without eliminating the EAC. This was not particularly disadvantageous for electrode fixation. The active electrode inserted into the cochlea was relatively stable because its intrinsic shape is modiolus-hugging and it could even be fixed with the aforementioned cochlear

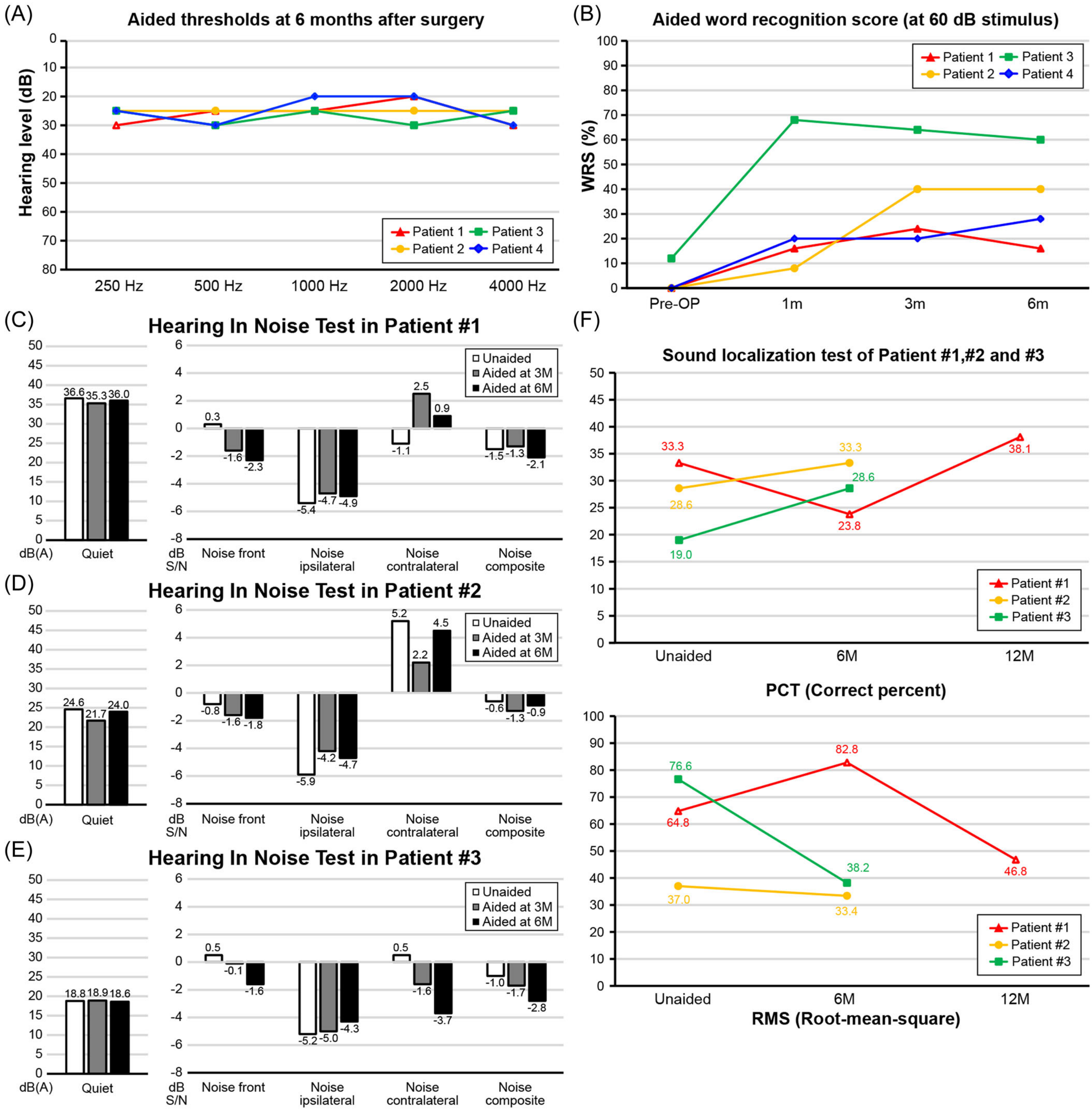

Figure 4. (A) Functional gain test after CI. (B) Aided word recognition scores for monosyllabic stimuli at 60 dB. (C–F) Results of the Hearing in Noise Test and sound localization test for patients #1 to #3.

reconstruction procedure. This procedure was particularly important for holding the placed electrode array. Electrodes could be fixed within the mastoid cavity in the same way as in conventional CI. As a result, it is acceptable to leave the EAC in place, provided that visibility can be secured for tumor removal and electrode insertion.

The duration of hearing loss and magnitude of tumor cochlear involvement affected the patient prognosis in this study. In previous studies, tumor size and extension were major factors affecting the ability to preserve auditory nerve function after translabyrinthine schwannoma removal.[29] The preoperative pure-tone threshold increased in patients with long durations of hearing loss and large lesions. In addition, the lesion size was proportional to the duration of hearing loss. For example, patient #1 had a 10-year duration of hearing loss and a tumor extending from the basal turn to the apical turn, whereas patients #3 and #4 had 2- to 3-year durations of hearing loss and lesions limited to the basal and middle turns, respectively. This pattern was also observed in a previous study.[29] Tumor extension, a long duration of hearing loss, and preoperative hearing are the most important factors affecting audiological outcomes after intracochlear schwannoma removal and CI.

Conclusions

If the modiolus is not completely invaded by an intracochlear schwannoma, simultaneous CI and tumor removal can be performed successfully. A slim modiolar electrode was placed at the modiolus after intracochlear schwannoma removal. The surgery resulted in good hearing performance, and lesion size and the duration of hearing loss were major factors affecting postoperative hearing outcomes.

Author Contributions

Jungho Ha, conception and design, acquisition and analysis of data, writing and critical revision of manuscript, final approval, accountability agreement; **Hantai Kim**, conception and design, acquisition and analysis of data, critical revision of the manuscript, final approval, accountability agreement; **Ga Young Gu**, conception and design, analysis and interpretation of data, critical revision of the manuscript, final approval, accountability agreement; **Young Jae Song**, conception and design, analysis and interpretation of data, critical revision of the manuscript, final approval, accountability agreement; **Jeong Hun Jang**, conception and design, interpretation of data, critical revision of the manuscript, final approval, accountability agreement; **Hun Yi Park**, conception and design, analysis and interpretation of data, critical revision of the manuscript, final approval, accountability agreement; **Yun-Hoon Choung**, conception and design, analysis and interpretation of data, critical revision of the manuscript for important intellectual content, final approval of the submitted version, accountability agreement.

Disclosures

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Supplemental Material

Additional supporting information is available in the online version of the article.

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