

Vestibular Schwannoma Resection with Ipsilateral Simultaneous Cochlear Implantation in Patients with Normal Contralateral Hearing

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Key Words

Vestibular schwannoma · Acoustic neuroma · Simultaneous implantation · Cochlear implant · Translabyrinthine approach · Single-sided deafness · Normal contralateral hearing

Abstract

Objective: To report the hearing results of cochlear implantation simultaneous to vestibular schwannoma (VS) resection by means of a translabyrinthine approach in patients with normal contralateral hearing. **Methods:** This was a prospective study including adults with sporadic VS. Tumors were resected by means of a modified translabyrinthine approach with preservation of the cochlear nerve. **Results:** A total of 13 patients underwent cochlear implantation. At 14 months, the mean pure-tone audiogram was 56 dB. The mean speech recognition was 80%. Cochlear implantation provides monaural and binaural benefits in all the conditions tested, including sound localization. **Conclusions:** Cochlear implantation can be safely performed simultaneously to VS resection with satisfactory hearing results provided that the cochlear nerve is anatomically intact.

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Introduction

Vestibular schwannoma (VS) patients represent a challenge both from surgical and audiological points of view. Whichever modality of management is chosen, the chances of preserving serviceable hearing in the affected ear in the long term are small [Tveiten et al., 2015]. Even with hearing preservation surgery, the chances of maintaining social serviceable hearing in the short term range from 0 to 37% in the most experienced hands [Rabelo de Freitas et al., 2012]. Patients with neurofibromatosis type 2 (NF-2) or VS in the only or better hearing ear deserve separate consideration, as they are at special risk of developing bilateral anacusia, either as a consequence of treatment or from the natural course of the disease [Vincenti et al., 2008; Celis-Aguilar et al., 2012; Lassaletta et al., 2016].

In the case of sporadic VS with normal contralateral hearing, the audiological impact of this disease focuses on the loss of binaural hearing. This supposes a diminished binaural summation effect (advantage of hearing with identical signal arriving at both ears), a reduced squelch effect (ability of the brain to separate noise and speech when both arrive from different locations), and problems

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linked to the head shadow effect (diminished speech understanding when the head is between the source of the speech and the hearing ear). In addition, these patients have a reduced ability to localize sound and speech [Vermeire and Van de Heyning, 2009; Kamal et al., 2012].

It is known from previous studies that patients with sporadic VS in the only or better hearing ear benefit from cochlear implantation simultaneously to VS resection. However, their results do not reach those of standard postlingually deaf patients in the majority of cases [Arriaga and Marks, 1995; Aristegui and Denia, 2005; Zannetti et al., 2008; Di Lella et al., 2013; Lassaletta et al., 2016]. In addition, little is known about their benefits in the presence of normal contralateral hearing.

To date, the cochlear implant (CI) is the only device capable of restoring true binaural hearing in profoundly deafened patients [Arndt et al., 2011]. Previous studies have reported moderate binaural benefits with cochlear implantation in standard postlingually single-sided deaf (SSD) patients [Van de Heyning et al., 2008; Vermeire and Van de Heyning, 2009; Arts et al., 2012]. However, it is doubtful whether these benefits would be noticeable in patients with VS, where it is known that CI offer less performance than in standard deafened patients.

Furthermore, it is still not clear how the integration of electrical hearing and natural hearing in the central nervous system is perceived in these patients. Hassepass et al. [2016] have recently reported a hearing benefit for ipsilateral implantation after resection of VS in SSD cases. These observations, still worth mentioning, represent only isolated cases, and implantation was performed not simultaneously, but 1 year after surgery. To our knowledge, there are no current published studies on cochlear implantation results performed simultaneously to VS resection for series of patients with SSD.

The purpose of this prospective study was therefore to assess the hearing results and subjective benefits of cochlear implantation simultaneously to tumor resection in sporadic VS patients when contralateral hearing is normal. Our objective was to evaluate the CI in monaural conditions but also the binaural benefit perceived by the patient.

Methods

This prospective study was conducted according to the principles stated in the Declaration of Helsinki.

Recruitment

Thirteen patients affected with sporadic VS were consecutively recruited for this study. There was evidence of growth in all the tu-

mors. In every case, there were one or more factors to advise against hearing preservation surgery [Sanna et al., 2004]. These factors were the following: (1) completely occupied internal auditory canal fundus, defined as absence of liquid in T2-enhanced magnetic resonance (MRI) sequences, (2) excessive dilatation or enlargement of the internal auditory canal, defined as a minimum of 50% increase in the anteroposterior diameter with respect to the contralateral side, (3) age greater than 65 years in candidates for the middle fossa approach. Patients were informed about the chances of preserving their hearing in our center in their specific circumstances [Rabelo de Freitas et al., 2012], and those who chose the translabyrinthine approach with a simultaneous CI were recruited for the study.

Inclusion criteria were as follows: patients with VS and socially useful bilateral hearing (classes A and B of the modified Sanna classification) [Kanzaki et al., 2003], tumor size intrameatal or class I of the Kanzaki classification [Kanzaki et al., 2003], aged over 18, and able to understand the potential risks and benefits of the procedure and its alternatives.

Intraoperatively, patients were excluded according to the surgeon's criteria if there had been damage to the acoustic nerve evidenced by examination under the microscope. In addition, those patients with complete anatomical preservation of the cochlear nerve, but in whom excessive retraction, manipulation or elongation had been unavoidable for safe resection of the tumor, were excluded from the study and therefore did not undergo cochlear implantation.

Patients were extensively counseled in order to provide realistic expectations. All patients were advised about the possibility of damage to the cochlear nerve during surgery, which would have led to aborting CI placement. They were also realistically informed about the current results of cochlear implantation in VS and their special circumstances of normal contralateral hearing.

Surgery

VS resection was performed by a modified translabyrinthine approach in all cases in combination with a middle-ear exclusion [Free et al., 2013; Polo et al., 2016] to reduce the risk of cerebrospinal fluid leak and meningitis due to CI placement. The surgical incision used was an inverted S shape with superior extension [Sanna, 2011]. All the patients were implanted in the same stage with Cochlear Nucleus Freedom implants (Cochlear Ltd., Lane Cove, N.S.W., Australia). Full electrode insertion into the scala tympani was achieved through the round window in all cases. On the second postoperative day, a control X-ray confirmed the correct positioning of the intracochlear electrodes. CI activation was performed on the first postoperative follow-up visit 4 weeks later.

Follow-Up

Patients were scheduled for follow-up every 3 months during the first year after surgery, every 6 months during the second year, and annually thereafter. Audiological tests were made and subjective benefit questionnaires filled in at 1, 6 and 12 months after activation and annually thereafter.

Hearing Tests

Monoaural CI Benefit. A pure-tone audiogram for the CI was obtained for the frequencies 250, 500, 1,000, 2,000, 4,000 and 8,000 Hz. Postoperative auditory performances were assessed in both closed-set (vowel identification, VI) and open-set (disyllabic word recognition, sentence recognition, common phrases comprehen-

sion) formats with a monitored live voice through the sound field at a level of 70 dB SPL. The Bocca and Pellegrini sentence list was utilized. Patients were tested in the free field condition. Contralateral hearing was masked with white noise according to the patient's hearing thresholds.

Binaural Benefit. Speech recognition in noise was tested in three different spatial configurations with 4 loudspeakers at the level of the subject's head at a distance of 1 m: (1) summation effect: speech and noise signal presented from the front (S_0N_0); (2) head shadow effect: speech presented from the CI side, noise presented from the front ($S_{CI}N_0$); (3) squelch effect: speech presented from the front, noise presented from the CI side (S_0N_{CI}). Cocktail party noise was presented at a constant level of 65 dB SPL. Consecutive lists of 10 disyllabic words were presented to the subject. The initial speech level was 65 dB; after each set of 10 words, the number of correct answers was recorded and the speech level adjusted in steps of 1 dB according to the subject's answer. Lists were consecutively presented until obtaining the signal-to-noise ratio at 50% of speech recognition. This binaural testing is detailed elsewhere [Vermeire and Van de Heyning, 2009].

Sound Localization. Four loudspeakers were located at intervals of 90° at the level of the subject's head at a distance of 1 m from the head at positions azimuth 0, 90, 180 and 270°. Two sets of 20 disyllabic words were randomly delivered from the speakers, in aided and unaided conditions, and the subject pointed to the source of the sound. The number of correct answers in each condition was recorded.

Benefit Questionnaires

To evaluate the subjective improvement with the CI, a set of questionnaires was administered at each visit:

(1) The Bern Benefit in Single-Sided Deafness Questionnaire consists of 10 visual analog scales rating the subjectively perceived benefit of the device in different situations [Kompis et al., 2011] (see online suppl. material 1; for all online suppl. material, see www.karger.com/doi/10.1159/000448583).

(2) The Single-Sided Deafness Questionnaire [Wazen et al., 2003] (see online suppl. material 2) consists of 5 items; the first two evaluate the frequency of use of the device, the third and fourth evaluate the general improvement in the quality of life and the satisfaction of the subject, and the fifth item evaluates the benefit of the device in 5 different daily situations.

Statistical Analysis

Data were analyzed with a statistical software program (SPSS Statistics for Windows version 20.0, Chicago, Ill., USA). Continuous data were summarized as median \pm interquartile range. Categorical data were presented as frequency counts and percentages. Univariate nonparametric tests for continuous dependent variables included the Wilcoxon test. Results with an alpha value equal or lower than 0.05 were considered statistically significant.

Results

Patients

Sixty-three patients were offered participation in this study according to the previously stated inclusion criteria.

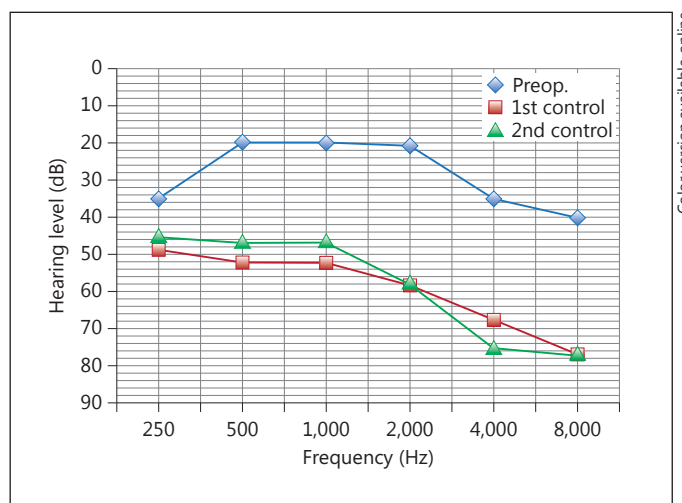


Fig. 1. Pure-tone audiogram showing CI performance in the monaural condition (masking on the normal-hearing side) preoperatively (preop.) and at 6 and 12 months.

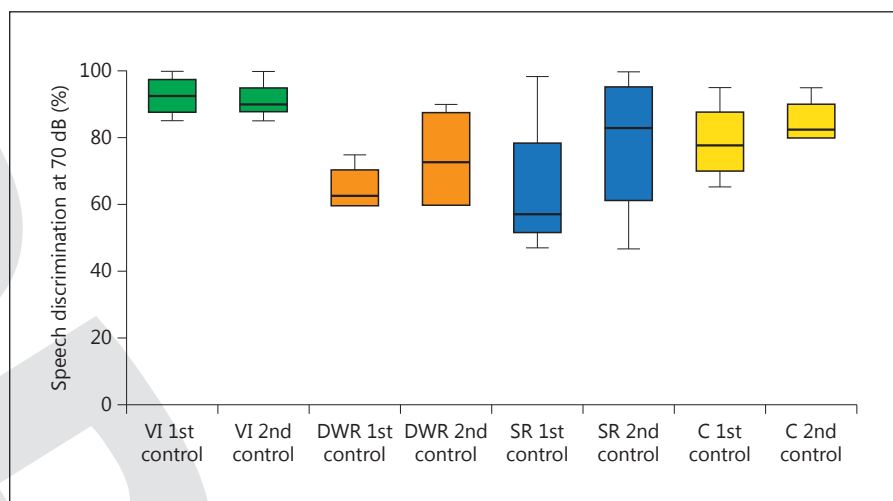
ria. Of these, 19 patients accepted participation in the study. Intraoperatively, the cochlear nerve was intentionally cut due to an unacceptable risk concerning the facial nerve during tumoral dissection in 3 patients. Furthermore, the cochlear nerve was not anatomically intact at the end of the procedure in 2 patients. In addition, in 1 case the tumor originated from the cochlear nerve. Thus, a total of 13 patients underwent cochlear implantation; the median age was 50 years (range 33–72 years). There were 8 males and 5 females. There were no complications in the immediate postoperative period. One patient had a delayed activation due to the presence of a late postoperative subcutaneous serohematoma, which was drained with a syringe, and a head bandage was applied.

Tumor size was class I in 2 cases and intrameatal in the remaining patients. All patients had ipsilateral preoperative hearing before surgery, therefore there was no deprivation time for these implants. Contralateral hearing was class A in all except 1 patient, in whom it was class B.

Patient Losses

At the time of collecting data for this study, 12 patients had completed the first control (6 months), and 5 patients had completed the second control (14 months). Due to the characteristics of our center, patients travel from all over the country and beyond, and they usually tend to extend the time between their follow-up appointments.

Fig. 2. Box-plot diagram showing vowel identification (VI), disyllabic word recognition (DWR), speech recognition (SR) and common phrases comprehension (C) at the first and second controls.



Therefore, the median time for the second control visit was 14 months (range 12–16 months).

One patient was unable to continue with the follow-up due to concomitant tumoral disease (no hearing tests were performed). Two patients were lost to the second control visit. One of them (case No. 7) had 10 disabled electrodes due to nonauditory perception, and the remaining ones were enabled but with weak electrical current because of nonauditory side effects (cephalea and nausea). The other patient (case No. 3) was unsatisfied with the outcome and stopped using the CI after 9 months. He was readapted to a contralateral routing of signals hearing aid. For both cases results at 6 months were included in the study.

Hearing Results

Hearing results are presented for the first control visit (12 patients) and for the second control visit (5 patients). Results for CI in the monaural condition and results for binaural tests are provided.

CI Performance with Masking on the Normal-Hearing Side

All patients had auditory sensation; however, 2 patients had unsatisfactory results with a mean pure-tone average (PTA) of 90 and 91.25 dB, respectively (cases No. 3 and 7).

PTA on the implanted side preoperatively and for the first and second controls are shown in figure 1. A discrete improvement for the mean PTA was noted in low frequencies, which was not statistically significant ($p > 0.05$).

Mean results for speech discrimination are shown in figure 2. Likewise PTA, disyllabic word recognition, sentence recognition and common phrases comprehension improved at the second control, but this improvement was not statistically significant ($p > 0.05$). All patients had speech perception except patient No. 7, who scored 0% in all the evaluations. This patient, however, had a PTA of 48.75 dB. Two patients had poor results, obtaining only some benefit regarding VI (patient No. 3 with 55% of VI and patient No. 9 with 35% of VI).

Binaural Hearing Benefits

The binaural benefit for the first control can be observed in figure 3a. There was an improvement in the 3 spatial configurations tested that was not statistically significant. At the second control (fig. 3b) there was a considerable improvement in the 3 spatial configurations in relation to the first control. The binaural benefit for the squelch effect (S_0N_{CI}) was statistically significant.

Figure 4 shows the results of sound localization in the unaided condition compared with the CI in the first and second controls. There was a statistically significant improvement in both controls compared with the unaided condition ($p = 0.027$).

Subjective Questionnaires

Subjective questionnaires are presented for the first control (fig. 5–7). In the Bern Benefit for Single-Sided Deafness Questionnaire (fig. 5), all patients but 1 expressed benefit with the CI in the 10 daily situations questioned.

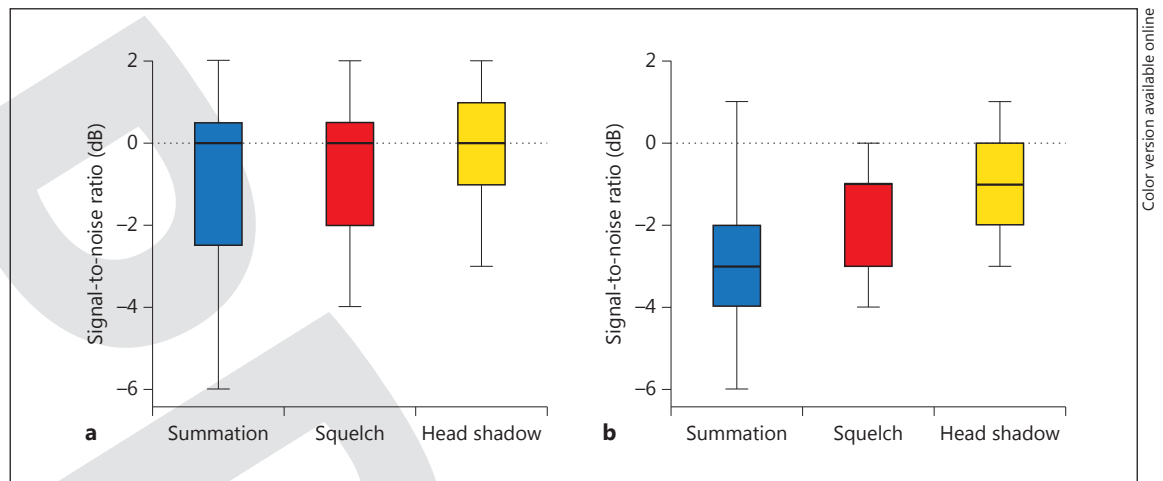


Fig. 3. Box-plot diagram showing the median changes in signal-to-noise ratio (difference between unaided and aided conditions) for the 3 conditions tested when the CI was connected. Values below the horizontal dotted line mean a decrease in signal-to-noise ratio, indicating speech comprehension improvement when the CI was connected. There is significant improvement between the unaided and aided conditions in the first ($p = 0.027$) and second controls ($p = 0.043$). **a** First control. **b** Second control.

The results for the Single-Sided Deafness Questionnaire are depicted in figures 6 and 7. Ninety percent of the patients reported using the CI between 5 and 7 days per week. Between 60 and 80% of the patients reported benefit in the 5 circumstances evaluated; no patient reported worsening with the CI. The median satisfaction score on a 10-point scale was 8 (fig. 7).

Discussion

Cochlear implantation has been previously described for the hearing rehabilitation of patients with NF-2 and sporadic VS in the only or better hearing ear [Lustig et al., 2006; Odat et al., 2011; Carlson et al., 2012; Di Lella et al., 2013; Lloyd et al., 2014; Aristegui and Denia, 2015; Lasaletta et al., 2016]. However, little is known about their benefits in the presence of normal contralateral hearing after VS resection.

This study has been able to prove that cochlear implantation can be safely performed simultaneously to VS resection with satisfactory hearing results provided that the cochlear nerve is anatomically intact and no aggressive maneuvers are applied to resect the tumor.

CI offered binaural benefit in all the conditions tested. Statistical significance was reached for spatial configuration No. 2 (squelch effect) and for sound localization. As mentioned in the introduction, the squelch binaural ben-

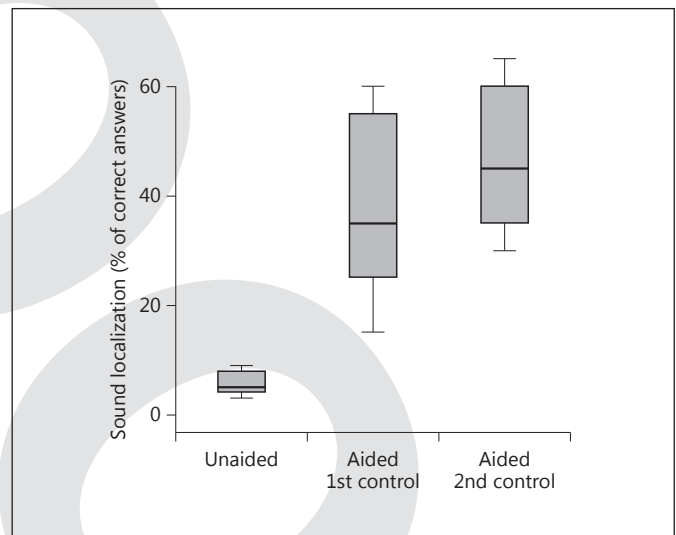
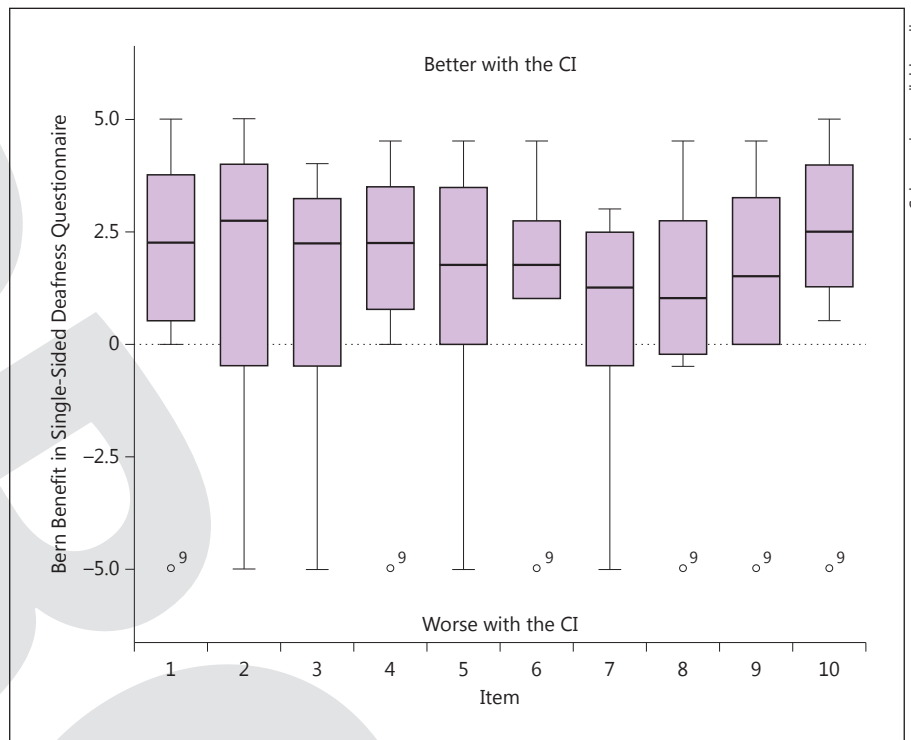


Fig. 4. Box-plot diagram showing sound localization in the unaided condition, and with the CI at the first and second controls.

efit refers to the improvement in speech perception when the signal and the noise come from different locations. In the same manner as sound localization, the central nervous system analyzes the interaural differences in the signal arriving at both ears, in order to separate the relevant information from the noise. Both sound localization and

Fig. 5. Diagram showing the scores of the Bern Benefit in Single-Sided Deafness Questionnaire. The majority of patients reported benefit in 10 daily situations when the implant was connected. 1 = To follow a conversation with one person in a quiet environment; 2 = to understand a TV or radio speaker; 3 = to listen to music; 4 = to follow a conversation from some distance (5 m/ 15 ft or more); 5 = to follow a conversation with background noise; 6 = to hold a conversation while driving a car; 7 = to understand speech in a reverberant room, such as a large entrance hall or a church; 8 = to participate in a conversation with 3 or more participants; 9 = to localize a sound source, such as a honking car; 10 = overall, hearing is easier with/without the CI.



squelch effect are based on higher-order processing and, in contrast to head shadow and summation effects, they depend on more complex signal processing based on the presence of 2 independent acoustic sensors [Kamal et al., 2012]. As addressed in the literature, devices such as contralateral routing of signals or bone-anchored hearing systems are capable of providing hearing benefit for summation and head shadow effect, whereas for squelch and sound localization, the CI is the only device providing benefit to date [Kamal et al., 2012]. This would explain the robustness of the benefit obtained with the CI for these 2 conditions in our study.

However, the clinical relevance of our findings may be called into question. As detailed in Methods, the main outcome measure of our binaural tests was changes in the speech-in-noise test. As Nilsson et al. [1994] pointed out, the shift of 1 dB in the speech-in-noise test corresponds to 10% change in speech discrimination. Hence, at the second control (fig. 3b) it can be appreciated that although the median improvement in the speech-in-noise test was between 1 and 3 dB, some patients obtained as much as 6 dB of improvement. Furthermore, these observations are supported by the positive evaluation provided by the patients on the subjective questionnaires (fig. 6, 7).

The results of this study must be interpreted with caution. Cochlear implantation has been demonstrated to progressively improve its results, stabilizing from the second year on. Since the mean follow-up for the second control was relatively short (14 months), these results may probably improve on a longer-term follow-up. In fact, the benefit obtained for the summation and the head shadow effect may become significant, as happened with the squelch effect at the second control.

The most controversial aspect of performing VS resection and CI placement simultaneously is the lack of reliable intraoperative monitoring of the eighth cochlear nerve to determine whether, after tumor resection, this nerve would be suitable to conduct electrical stimuli delivered by a CI. The distress to the cochlear nerve can be the consequence not only of the surgical maneuvers during tumor removal, but also of tumor-related factors such as compression of the cochlear nerve in the internal auditory canal or pressure of the tumor in the fundus of the internal auditory canal leading to irreversible loss of spiral ganglion cells.

Once it has been established that a CI is capable of providing good hearing results after VS resection, offering even monaural and binaural benefits, the efforts should be directed towards developing an intraoperative testing

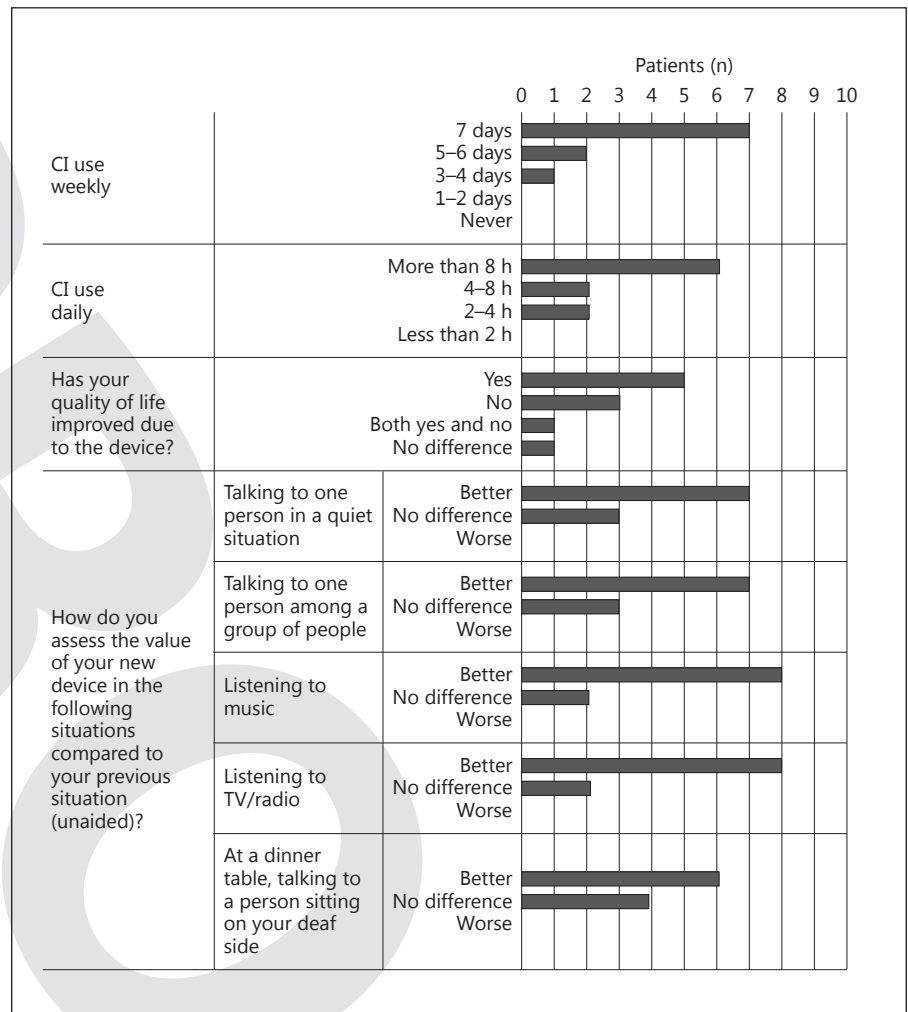


Fig. 6. Bar diagram showing the objective evaluation of the patient's CI according to the Single-Sided Deafness Questionnaire, items 1, 2, 3 and 5.

method capable of determining whether an intact cochlear nerve after a VS resection is or will be able to conduct impulses originating from a CI. The current promontory test or electrical stimuli applied to the promontory has been shown to be controversial for predicting CI results, as a negative response does not necessarily mean a lack of benefit from the CI [Kuo and Gibson, 1991, 2002] (needless to say it is even less reliable if the test is done immediately after VS resection).

There are some options already available on the market to try to obtain this information intraoperatively, prior to implantation, such as the dorsal cochlear nucleus action potential electrode Neuromaster® intraoperative monitoring system MEE-1100 (Nihon Kohden Ltd.), which consists of a needle stimulator which is directly applied to the cochlear nerve once the tumor is removed, and the answer is registered by a surface electrode in the

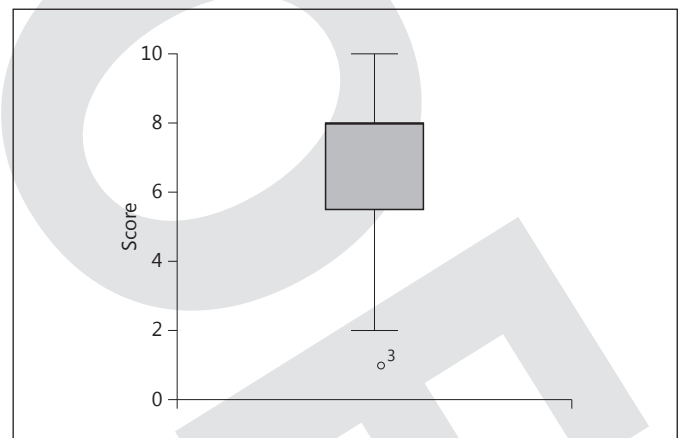


Fig. 7. Box-plot diagram showing the median score for item No. 4 of the Single-Sided Deafness Questionnaire: 'Try to determine your satisfaction or dissatisfaction with the device on a 10-point scale' (10 = very satisfied; 5 = no difference; 0 = unsatisfied).

cochlear nucleus [Magnan et al., 2013]. Some authors [Tysome et al., 2013] have also mentioned the possibility of an intracochlear electrode to obtain this information, although such a device is not currently standardized for clinical use. All of these devices have the same limitation. If the answer is positive, the chances of obtaining a good result with a CI may be high. On the other hand, if the answer is negative, it is impossible to know whether it is due to permanent cochlear nerve damage or a temporary contusion due to the surgery or tumoral compression. The results of this study rely on intraoperative patient selection based on the criterion of a very experienced surgeon. This, without any doubt, can affect the reproducibility of this study. In this regard, future studies will be needed to assess the correlation of intraoperative testing methods with CI performance and provide management recommendations for other clinicians.

Simultaneous tumor removal and cochlear implantation allows avoiding an additional major surgical procedure and permits initiating hearing rehabilitation from the very beginning.

Due to the fact that intraoperative testings are not optimal yet, if the decision of implanting is based exclusively on these tests, the chances of inserting a foreign device that could be useless are not negligible. Additionally, the economical debate must be taken into consideration. In this regard, it has been recently proposed to insert an intracochlear electrode at the time of the VS surgery to avoid cochlear ossification and to determine the cochlear nerve's functionality with a promontory test 12 months later, in order to proceed with implantation only in patients with viable cochlear nerves [Hasepass et al., 2016].

Another statement supporting delayed implantation is the possibility of performing a control MRI 1 year after tumor removal to rule out tumor recurrence before CI insertion [Hasepass et al., 2016]. However, our rate of tumor recurrence after complete tumor removal was extremely low, 0.05% [Ahmad et al., 2012]. Moreover, if there is reasonable suspicion of residual tumor or recurrence, there is the possibility of temporarily removing the magnet of the CI under local anesthesia to avoid distortion of the MRI image. Nevertheless, in cases of sporadic VS with normal contralateral hearing, if residual tumor is left intentionally or unintentionally, it is the recommendation of the author to abort CI placement, as oncological surveillance is prioritized. Patients with impaired contralateral hearing or NF-2 deserve separate consideration and can benefit from other strategies of management such as contralateral CI or CI without tumoral resection [Di Lella et al., 2013; Lassaletta et al., 2016].

Furthermore, patients operated on due to VS frequently refuse additional procedures and surgeries if these are not strictly related to their survival. In this regard, simultaneous implantation has clear advantages over delayed implantation. In addition, it may seem to provide better hearing outcomes than delayed implantation [Lassaletta et al., 2016], as it reduces the risk of intracochlear ossification and permits a more atraumatic surgery, eliminating the intermediate step of placing an intracochlear placeholder for several months. All of the factors mentioned above, plus the early onset of hearing rehabilitation, which is known to prevent spiral ganglion cell loss [Leake et al., 1999], could have contributed to the positive results evidenced in this study.

Strengths and Limitations

We would like to review the primary strengths and limitations of the study.

Our study represents the largest series of patients undergoing CI insertion simultaneously to sporadic VS removal. Moreover, it is the only existing series of this kind of patients with normal contralateral hearing.

Despite a close follow-up, 3 of our patients were unable to continue with the hearing evaluation. Two of them had poor results with the CI. The loss of these patients for the second control may have positively influenced the overall results for the second control. On the other hand, it is worth mentioning that the statistical analysis comparing the first and second controls has automatically excluded those patients who had not completed 12 months of follow-up. Thus, the results and conclusions are highly valid to patients who undergo the same management strategy.

The decision of placing the CI was made upon the surgeon's perception of cochlear nerve integrity under microscope examination. No intraoperative tests were used to assess cochlear nerve functionality. However, we tried to minimize external factors such as tumor size or preoperative hearing by applying relatively tight patient inclusion criteria. Nonetheless, further studies with intraoperative monitoring are needed in order to obtain relevant data for future clinical applications.

Conclusions

This is the largest series on cochlear implantation simultaneous to sporadic VS removal and the only one on cochlear implantation simultaneous to sporadic VS removal in single-sided deafness.

This study has been able to prove that cochlear implantation can be safely performed simultaneously to VS resection with satisfactory hearing results provided that the cochlear nerve is anatomically intact and no aggressive maneuvers are applied to resect the tumor. The CI provides monaural and binaural benefit in all the conditions tested, including sound localization. Simultaneous implantation has clear advantages over delayed insertion, therefore every effort should be made to implant simul-

taneously to tumor removal. Future investigations should be directed towards developing intraoperative testing to determine cochlear nerve functionality and therefore guide the decision of implanting or not.

Disclosure Statement

The authors declare no conflicts of interest.

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