The Laryngoscope

Long-term Audiologic Outcomes After Cochlear Implantation for Single-Sided Deafness

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Objectives: To evaluate the long-term audiometric outcomes, sound localization abilities, binaural benefits, and tinnitus assessment of subjects with cochlear implant (CI) after a diagnosis of unilateral severe-to-profound hearing loss.

Method: The study group consisted of 60 (mean age 52 years, range 19–84) subjects with profound hearing loss in one ear and normal to near-normal hearing in the other ear who underwent CI. Data analysis included pre- and postoperative Consonant-Nucleus-Consonant (CNC) Word scores, AzBio Sentence scores, pure tone thresholds, sound localization, and Iowa Tinnitus Handicap Questionnaire scores.

Results: Preoperative average duration of deafness was 3.69 years (standard deviation 4.31), with an average follow-up time of 37.9 months (range 1–87). CNC and AzBio scores significantly improved (both $P < 0.001$) postoperatively among the entire cohort, and there was much heterogeneity in outcomes with respect to deafness etiology subgroup analysis. Sound localization abilities tended to improve longitudinally in the entire cohort. Binaural benefits using an adaptive Hearing in Noise Test test showed a significant ($P < 0.001$) improvement with head shadow effect. Utilizing the Iowa Tinnitus Handicap Questionnaire, there was significant improvement in social, physical, and emotional well-being ($P = 0.011$), along with hearing abilities ($P = 0.001$).

Conclusions: This case series is the largest cohort of CI SSD subjects to date and systematically analyzes their functional outcomes. Subjects have meaningful improvement in word understanding, and sound localization tends to gradually improve over time. Binaural benefit analysis showed significant improvement with head shadow effect, which likely provides ease of listening.

Key Words: Cochlear implants, sensorineural hearing loss, quality of life.

Level of Evidence: 4

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INTRODUCTION

Since 1961, cochlear implants (CI) have been able to restore hearing in deafened individuals by replacing function of an impaired inner ear in individuals with bilateral severe-to-profound hearing loss.[1–3] With advancements in technology and the benefits of improved speech understanding in quiet and noise, better sound localization, and overall quality of life seen with electrical stimulation,[4–7] the potential to expand implant criteria has profound implications. Subjects with unilateral severe-to-profound hearing loss and normal to near-normal hearing in the contralateral ear, also known as single-sided deafness (SSD), have the potential to meaningfully benefit from a CI.

SSD has profound auditory and psychosocial implications, including decreased ability for sound localization, binaural summation, binaural squelch effect, and quality of life.[8,9] The ability to localize sound with unilateral hearing loss is diminished due to the absence of interaural timing and intensity differences between the two hearing ears.[9,10] Historically, SSD has been treated with a Contralateral Routing of the Signal (CROS) hearing aid or an osseointegrated implant.[9,11] However, literature has showed little benefit of these devices with respect to speech perception in noise and sound localization.[12] Additionally, subjects with SSD commonly experience tinnitus, and electrical stimulation via a CI provides a possibility to suppress tinnitus.[13–16]

Previous studies have assessed early audiometric outcomes after CI for SSD.[7–9,17–22] Preliminary results have shown that CIs restore auditory function in a deafened ear, with improvement in speech recognition in noise, sound localization, and tinnitus control.[7,9,19,20] In a recent analysis of 34 subjects with SSD with 60 months of follow-up, Távora et al. showed that CIs improve speech perception in noise and sound localization, but tinnitus outcomes were not assessed.[23] Mertens et al. showed that quality of hearing was stable after 36 months, and speech perception in noise continued to improve at 36 months in

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an analysis of 22 patients.[18] However, most studies have been small case series with short-term follow-up. Whether these findings translate long-term is an active area of investigation.

SSD patients differ significantly from patients with severe-to-profound sensorineural hearing loss (SNHL) in that they have normal or near normal acoustic hearing in the contralateral ear. Thus, the central neural adaptations that occur following CI for patients with SSD may differ both temporally and functionally from what occurs in patients with bilateral hearing loss. Longitudinal data are needed to better understand how and to what extent patients with SSD integrate an acoustic signal from the normal ear with a CI signal over time. To better elucidate the long-term outcomes of cochlear implantation in SSD subjects, the aim of the current study was to examine the audiometric outcomes, sound localization abilities, tinnitus changes, and binaural benefits in a longitudinal fashion.

MATERIALS AND METHODS

Subjects

Institutional review board approval at the University of Iowa (Iowa City, IA) was obtained for this study. A portion of these demographics and initial results have previously been published.[9] Inclusion criteria for the current study was an expanded cohort of subjects with a history of unilateral profound hearing loss in one ear (referred to as CI ear) and normal to good audiometric profile (referred to as NH ear) in the opposite ear. Preoperative imaging is routinely completed to assess cochlear abnormalities and anatomical variations that could influence surgical candidacy. Preoperative imaging was completed in 58 (96.7%) subjects in the originally collected database, and two subjects did not have imaging recorded in the database. Implant activation typically occurs 1 month following surgery. Subjects were excluded if they were lost to follow-up. From 2011 to 2017, 60 subjects received a CI at our institution who met inclusion criteria. Demographic, audiometric, radiographic, and outcomes data were extracted from individual medical records and tabulated into a database.

Speech Perception and Sound Localization

Audiometric analysis took place preoperatively; postoperatively at 3, 6, and 12 months; and annually. Testing was conducted in a double-walled booth and was performed by a CI audiologist. Methods for sound localization and speech perception have previously been described.[9] Briefly, speech perception in quiet was measured in the sound field with Consonant-Nucleus-Consonant words (CNC) and AzBio sentences[24,25] with direct connect using a subjective most comfortable level. The Hearing in Noise Test was administered with sentences always presented at 0°. Noise was alternated from 0°, 90° facing the CI ear, and 90° facing the NH ear. In all conditions, the competing noise was presented at a constant loudness of 65 dBA of sound pressure level (SPL). The loudness of the sentences presented was adapted throughout the test depending on whether the subject repeated the sentence correctly. A signal-to-noise ratio (SNR) was calculated based on how loud the sentences were presented above the noise floor for the subject to repeat them correctly 75% of the time. Presentation in each noise condition was tested in sound field with NH ear and in the everyday listening condition (NH ear and CI ear together). Sound localization was completed at a threshold of 60 dBA SPL using 16 different everyday sounds, which were presented randomly from eight speakers arranged in a 108° horizontal arc. A sound was played 12 times from each loudspeaker for a total of 96 presentations.[26] Preoperatively, a hearing aid was utilized in the worst hearing ear if the specific subject's hearing loss permitted. Postoperatively, the subject was tested using the everyday listening condition. The subject was instructed to identify which speaker the sound originated from but to not identify the specific sound. Localization scores were calculated utilizing the average root-mean-

TABLE I.
Summary of Clinical, Demographic, and Implant Characteristics.

	n = 60
Mean age at implant (range), years	52 (19–84)
Gender, n (%)	
Female	27 (45%)
Male	33 (55%)
Duration of deafness, years (SD)	3.69 (4.31)
Preoperative hearing	
AzBio score (mean, SD)	5 (12)
CNC score (mean, SD)	8.4 (19)
PTA (mean, SD)	90 (21.4)
WRS (mean, SD)	17 (17.6)
Cochlear implant type	
Advanced Bionics Mid-Scala	3 (5%)
Clarion Hi-Res 90K	14 (23.3%)
Clarion Hi-Res 90K Mid-Scala	1 (1.67%)
Med-EL Flex 24	4 (6.7%)
Med-EL Flex 28	1 (1.67%)
Nucleus 512	5 (8.3%)
Nucleus CI 24RE	7 (11.67%)
Nucleus CI422	21 (35%)
Nucleus CI522	2 (3.3%)
Nucleus CI532	2 (3.3%)

CNC = consonant nucleus consonant test; PTA = pure tone average; SD = standard deviation; WRS = word recognition score.

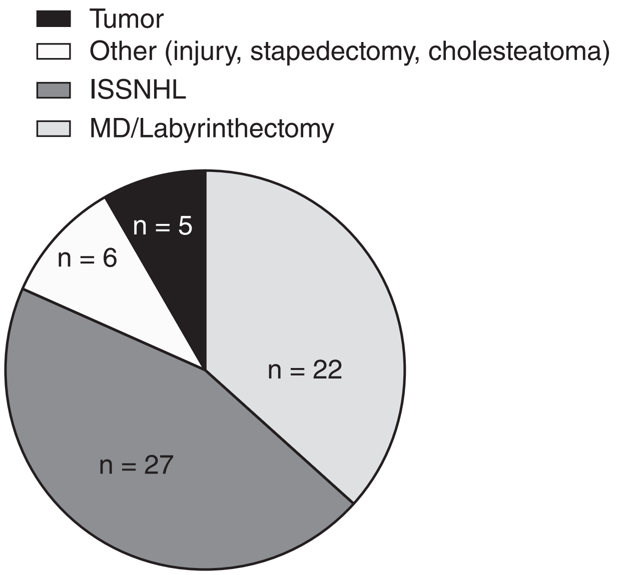
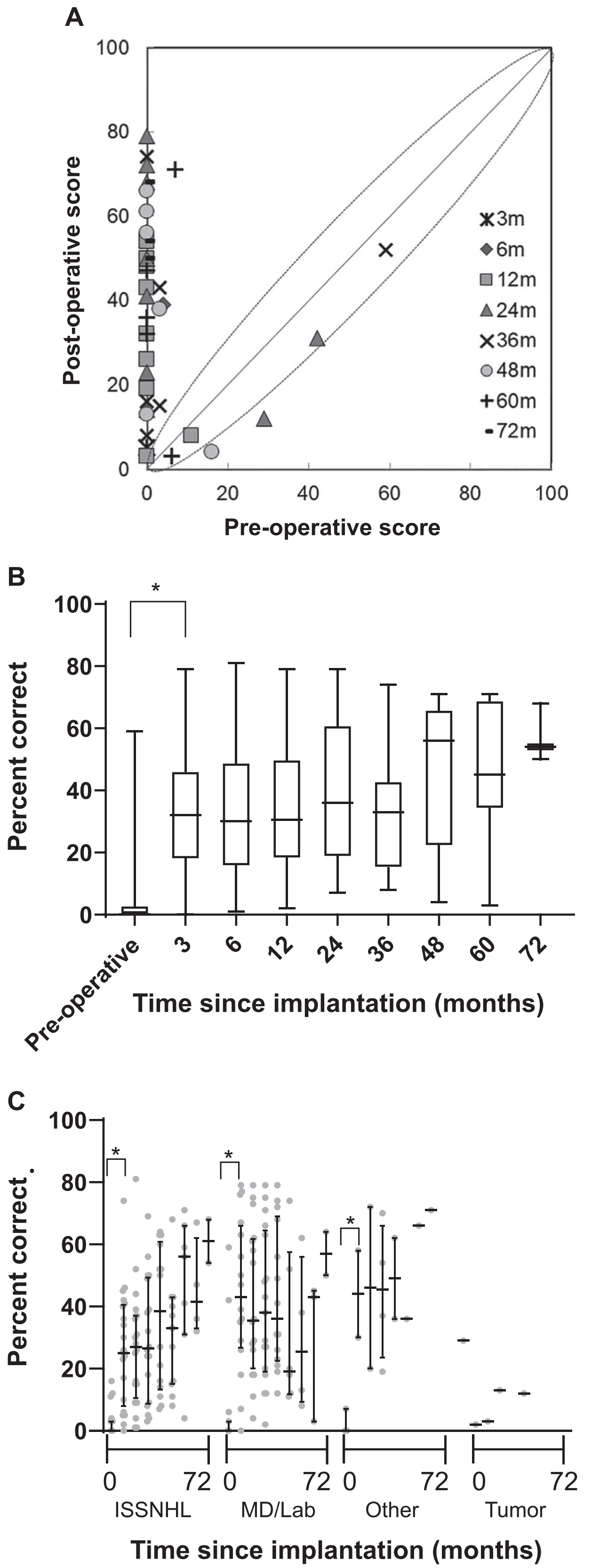


Fig. 1. Preoperative diagnosis of cause of single-sided deafness in the study cohort.

squared (RMS) error in degrees.[26] Pure tone audiometry (PTA) and word recognition scores (WRS) were assessed on the operated and nonoperated surgical side preoperatively.



Tinnitus

For tinnitus assessment, the Iowa Tinnitus Handicap Questionnaire, a psychometrically validated tool consisting of 27 questions,[27] was administered to subjects at pre- and postoperative follow-up at annual visits. The questionnaire assesses how a subject's life is affected by tinnitus and also may be used to monitor for changes in tinnitus after implantation of a CI. Factor 1 reflects subjects' social, emotional, and physical behaviors; factor 2 assesses hearing ability; and factor 3 represents their view on tinnitus.[27]

Statistical Analysis

For univariate analysis, Kruskal-Wallis testing was used to compare pre- and postoperative audiometric scores. Correlation of sound localization changes over time compared to baseline measurements were investigated with Spearman correlation. Speech perception scores were measured as percent correct. All tests were considered statistically significant with $P < 0.05$. Statistical analysis was performed using SAS 9.4 (SAS Institute Inc., Cary, NC).

RESULTS

Demographic, clinical, and surgical implant characteristics are summarized in Table I. The study population was comprised of 33 (55%) males and 27 (45%) females, with an average age of 52 years (range 19–84). The average length of follow-up with one of the senior surgeons or audiologists was 37.9 months (range 1–87). The average preoperative AzBio score was 5 (SD 12); CNC was 8.4 (SD 19); and WRS was 17 (SD 17.6). The average audiogram $PTA_{500:\ 1000;\ 2000;\ 4000\ Hz}$ of the CI ear was 90 dB (SD 21.4) and 23 (SD 13) of the NH ear. Twenty-four patients had used a hearing aid at some point prior to CI. There was significant heterogeneity in the type of CI used, with the Nucleus CI422 and Clarion Hi-Res 90K being the most common at 35% and 23%, respectively. The etiology of preoperative SSD is displayed in Figure 1. Idiopathic sudden sensorineural hearing loss (ISSNHL) (43%) was the most common etiology, followed by Ménière's disease (35%). All the patients with Ménière's disease underwent simultaneous labyrinthectomy for the control of recalcitrant vertigo attacks and cochlear implantation, as previously described.[9] Thus, their preoperative hearing status, although poor, was better than it would be postoperatively without use of a CI. One subject had sudden SNHL hearing loss due to labyrinthitis 3 weeks after a stapedectomy for otosclerosis and was implanted ~4 months after the sudden SNHL. All patients were evaluated 3 months postoperatively, 43 patients at 6 months, 41 patients at

Fig. 2. CNC word outcomes in patients with CI for SSD. (A) Individual scatter plot of preoperative CNC words in quiet in the implanted ear only compared with the most recent postoperative score. (B) Box-and-whisker plot of entire cohort CNC scores in quiet over time since implantation. Three months postoperative CNC score significantly improved ($P < 0.001$, Kruskal-Wallis test) compared to preoperative score. CNC scores did not significantly ($P = 0.16$) improve after the initial 3-month follow-up period. C. Pre- and postoperative CNC scores in quiet stratified by preoperative SSD etiology. All subgroups except the tumor cohort significantly ($P < 0.05$, Kruskal-Wallis test) improved postoperatively.
CI = cochlear implant; CNC = consonant-nucleus-consonant; ISSNHL = idiopathic sudden sensorineural hearing loss; MD = Ménière's disease; SSD = single-sided deafness.

12 months, 35 patients at 24 months, 19 patients at 36 months, 14 at 48 months, 10 at 60 months, and 3 at 72 months. Four subjects no longer or rarely used their CI due to poor performance postactivation throughout our study period.

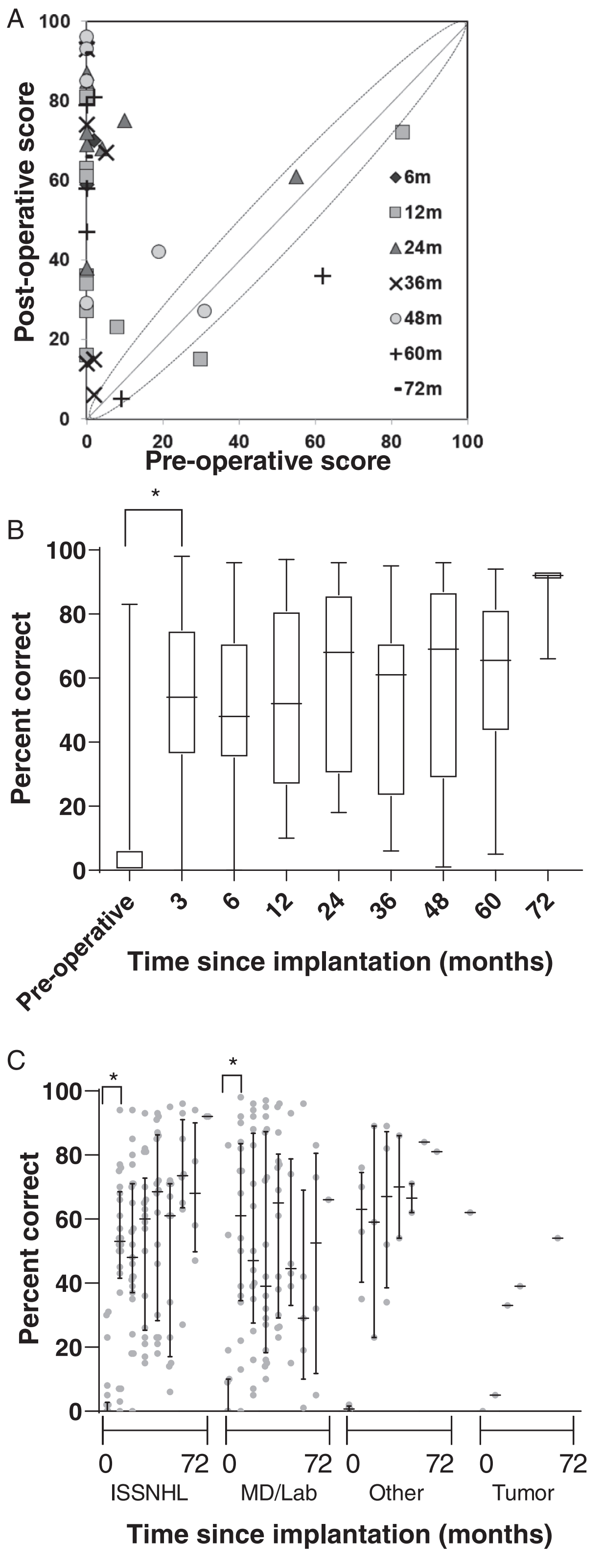


Fig. 3. AZBio sentence outcomes in patients with CI for SSD. A Individual scatter plot of pre-operative AzBio scores in quiet in the implanted ear only compared with the most recent post-operative score. B. Box and whisker plot of entire cohort AzBio scores over time since implantation. Three months post-operative AzBio score significantly improved ($P < 0.001$, Kruskal-Wallis test) compared to pre-operative score. AzBio scores did not significantly ($P = 0.11$) improve after the initial 3-month follow-up period. C Pre-and postoperative AzBio scores in quiet stratified by pre-operative SSD etiology. All sub-groups except the tumor cohort significantly ($P < 0.05$, Kruskal-Wallis test) improved post-operatively.
CNC = consonantnucleus-consonant; SSD = single-sided deafness; ISSNHL = idiopathic sudden sensorineural hearing loss; MD = Ménière's disease.

Pre- and postoperative CNC scores for the CI ear, which includes 72 months of follow-up in 3 subjects, are displayed in Figure 2. CNC scores significantly ($P < 0.001$) improved at the first audiometric follow-up visit, but there was no major change comparing further follow-up analysis ($P > 0.05$). There was a mean improvement of 42.3% at 3 months postoperatively. Subjects in the "other" cohort and ISSNHL improved the most with respect to their preoperative score. Similar pre- and postoperative AzBio scores are depicted in Figure 3. AzBio scores significantly ($P < 0.001$) improved at the first audiometric follow-up visit, but there was no major change comparing further follow-up analysis ($P > 0.05$). ISSNHL and subjects in the "other" cohort improved the most in AzBio scores with respect to their preoperative score.

Figure 3 displays sound localization results. Preoperatively, subjects with a history of Ménière's disease (RMS error 27.1°) had the best ability to localize sound, whereas subjects in the tumor subgroup had the worst (RMS error 45.5°). Although sound localization did not significantly ($P = 0.25$) improve at 3 months postoperatively compared to preoperative assessment in the entire cohort, sound localization tended to gradually improve at additional follow-up with time. Among subgroup analyses (Fig. 3C), subjects in the "other" cohort significantly ($P = 0.028$) improved their localization with time. Age, duration of deafness, preoperative PTA, preoperative WRS, change in CNC, and change in AzBio did not correlate with the change in sound localization between pre- and postoperative assessment ($P > 0.05$) (Supporting Table SI).

The benefits of binaural hearing with the addition of the CI were assessed and presented in Figure 4. The benefit was evaluated by comparing the SNR of the NH ear and the everyday listening condition. Binaural summation benefit was evaluated by comparing the NH ear only SNR to the everyday listening condition with the noise at 0° azimuth. Head shadow benefit was evaluated by comparing the NH ear only SNR to the everyday listening condition when the noise is at 90° azimuth facing the NH ear. Binaural squelch was assessed by comparing the NH ear only SNR to the everyday listening condition when the noise is at 90° azimuth facing the CI ear. On average, there was a significant improvement in head shadow effect ($P < 0.001$); however, there were no significant changes with respect to summation or the squelch effect ($P > 0.05$). There was significant heterogeneity seen in these categories with respect to subject's responses.

Results of the Iowa Tinnitus Handicap Questionnaire were used to assess subjects' emotional, social, and physical characteristics, along with the auditory abilities and perceptions (Fig. 5). Results from the most recent follow-up were used for assessment. Subjects in the ISSNHL group tended to have the best tinnitus results with respect to factors 1, 2, and 3, although no consistent trend was seen for analysis. When comparing tinnitus scores with duration of deafness, there was no trend that could be identified.

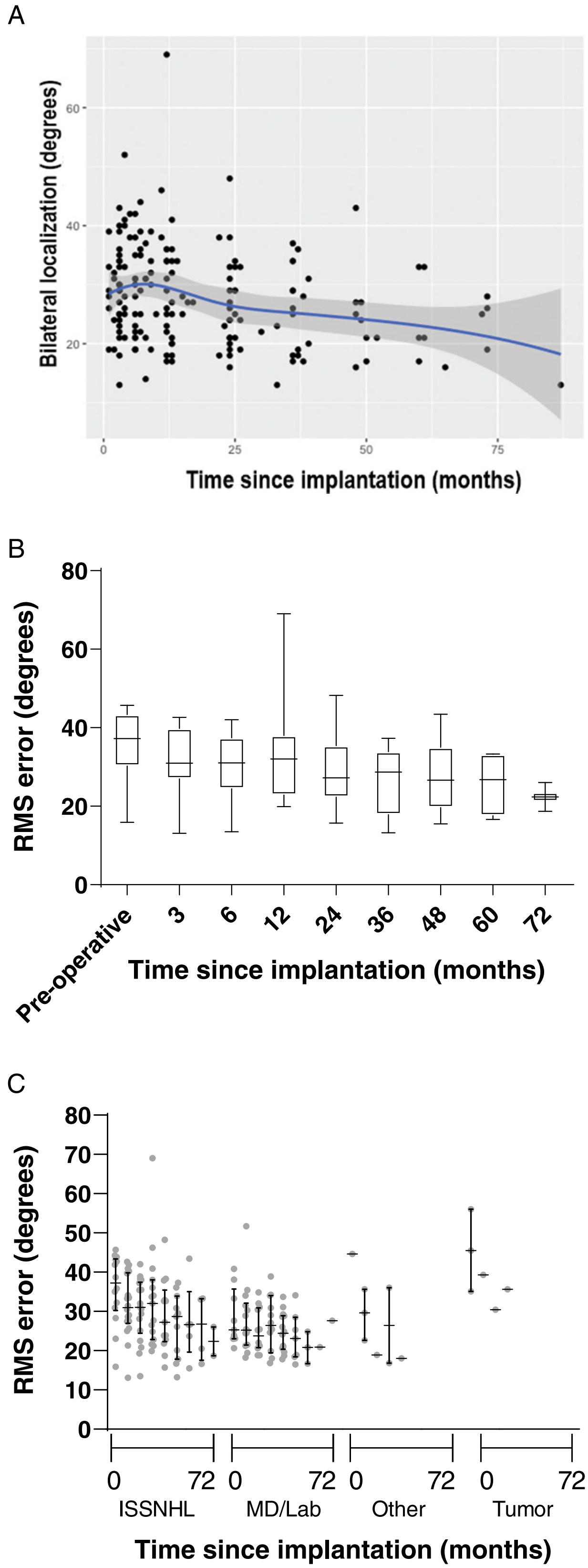


Fig. 4. Sound localization outcomes in patients with CI for SSD. (A) Individual scatter plot of patient ability to localize sound over time postoperatively. (B) Box-and-whisker plot of entire cohort bilateral sound localization over time. The cohort tended to improve localization abilities over time, although this did not reach statistical significance ($P = 0.25$, Kruskal-Wallis test). (C) Pre- and postoperative sound localization stratified by preoperative SSD etiology. Patients in the "other" subgroup significantly ($P = 0.028$, Kruskal-Wallis test) improved compared to preoperatively.
CI = cochlear implant; CNC = consonant-nucleus-consonant; ISSNHL = idiopathic sudden sensorineural hearing loss; MD = Ménière's disease; SSD = single-sided deafness. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

DISCUSSION

Historically, CROS hearing aids and osseointegrated implants have been the mainstay of treatment for subjects with SSD. The first report of a CI in SSD was by Van de Heyning et al. in 2008, which was intended to treat severe tinnitus and not necessarily provide binaural hearing.[14] In recent years, studies on the use of CI for SSD have demonstrated improvement in sound localization, speech in noise, quality of life, and tinnitus severity.[20,23] In an analysis of 34 SSD subjects treated with a CI, subjects showed an overall improvement in hearing abilities and reduction in listening efforts after a CI.[23] Despite these meaningful findings, cochlear implantation is not yet part of the routine management algorithm for subjects with SSD. This is likely multifactorial due to the lack of insurance coverage, initial increased costs compared to traditional hearing aids, and desire to avoid surgical implantation, among other factors.[22]

Significantly, the Food and Drug Administration recently approved Med-EL (Innsbruck, Austria) devices for SSD rehabilitation. Further, an ongoing randomized study aims to assess the cost-to-utility ratio between CI and conventional treatment options, which will likely provide important financial data regarding decision making for SSD.[28] Until now, most studies on CI in SSD patients have been limited to small patient cohorts, with limited follow-up and minimal long-term prognostic data. Thus, we hope to provide more support for CI in SSD subjects to provide future individuals the option of binaural hearing restoration. While our results were being prepared for publication, Távora-Vieira published a report of 34 subjects with SSD who showed long-term benefit with speech understanding, sound localization, and quality of life following CI, consistent with the findings presented here.[23]

In the current study, restoration of auditory function greatly improved after implantation for SSD, which is consistent with similar reports.[8,9,20] Subjects tended to have an immediate benefit postactivation, with variable improvement with subsequent follow-up. At 6 months after

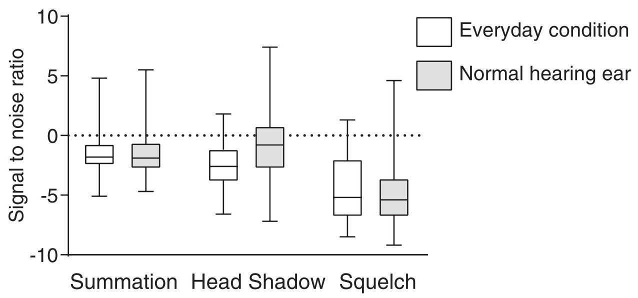


Fig. 5. Average of binaural benefits seen after CI for SSD. When noise was introduced to the listener in in the NH ear with the CI turned on (everyday condition), there was a significant ($P < 0.001$) improvement with the head shadow effect. No significant differences were seen with respect to binaural summation or the squelch effect. Nonimplanted is when sound is introduced with the CI turned off.
CI = cochlear implant; NH = normal to good audiometric profile; SSD = single-sided deafness is right.

implantation, our CNC average was 33%, which is less than subjects with a CI for binaural hearing loss, whereas they have a 61% CNC average.[29] These longitudinal results reveal that, although people with unilateral and bilateral hearing loss both benefit from implantation, there likely is a distinct multifactorial phenomenon occurring with rehabilitative auditory function among these unique groups that needs to be elucidated in future studies. Mertens et al. have suggested that an extended period of time may be necessary to integrate the electrical signal from a CI in subjects who have a normal contralateral hearing ear.[18]

The ability to localize sound is greatly compromised in the unilaterally deafened subject because binaural input is lost; however, a CI provides an effective route for restoration. Better sound localization is an additional benefit that is not possible in subjects with CROS hearing aids or osseointegrated implants.[9] In CI patients, previous reports have shown that localization improves but essentially

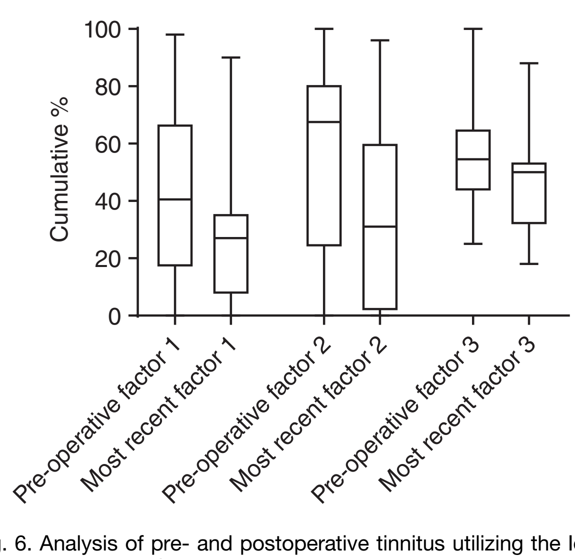


Fig. 6. Analysis of pre- and postoperative tinnitus utilizing the Iowa Tinnitus Handicap Questionnaire. Pre- and postoperative data was available for 28 patients. Factor 1 examines social, physical, and emotional well-being; factor 2 examines hearing abilities; and factor 3 examines an individual's view of tinnitus. Factor 1 ($P = 0.011$); factor 2 ($P = 0.001$); factor 3 ($P = 0.11$).

plateaus by less than 1 year after surgery.[11,30] Galvin et al. showed that the ability to localize did not significantly improve until at least 6 months postactivation.[20] The mean RMS error in our study was 23.35 at 60 months, whereas Távora-Vieira reported a RMS error of 24.6 in their study.[23] We demonstrate that sound localization tends to continue to improve throughout the postoperative period, and years of auditory rehabilitation with consistent CI usage may be necessary to see meaningful improvement. Additional long-term follow-up would be beneficial in assessing how much improvement is seen as our subjects continued to improve at the most recent assessment.

Individuals who lose binaural hearing subsequently develop difficulties with speech in noise and sound localization due to a deficit of binaural effects.[19] When assessing summation, squelch, and the head shadow effect, the most improvement was seen with a greater head shadow effect. These results are similar to Yoon et al., who showed that asymmetric stimulation among ears is least disrupted with the head shadow effect.[31] Currently, CROS hearing aids and osseointegrated implants address the head shadow effect; however, binaural hearing is still deficient because auditory stimulation is routed to the normal hearing ear.[8] Arndt et al. reported that SSD subjects demonstrate improved sound localization and speech comprehension when using a CI compared to CROS or osseointegrated implant.[17] Thus, CI for SSD subjects may be the only treatment modality to date that truly stimulates the deafened ear.

Tinnitus can be a debilitating associated symptom in patients with SSD or binaural hearing loss that has the potential to profoundly impact quality of life. Most tinnitus is subjective in that the patient is the only one who hears the noise.[14] Many hypotheses exist for the cause of tinnitus, including generation of tinnitus in the setting of auditory deprivation, as seen with SSD patients.[14] Stimulation of the auditory cortex with a CI may have the potential to ameliorate this troublesome side effect.[14] Previous reports have analyzed early outcomes of tinnitus after CI for SSD.[14,17] Arndt et al. demonstrated a significant improvement or complete resolution of tinnitus in eight out of 10 subjects.[17] We found that subjects' social, physical, and emotional well-being; hearing abilities; and perceptions of tinnitus all tended to improve after a CI. Given that there can be significant heterogeneity with respect to the severity and quality of tinnitus between subjects, the response and improvement seen with tinnitus scores is less easily predictable compared to objective audiometric outcomes. The results in the current study are encouraging for subjects with tinnitus associated with SSD; we have shown subjects are able to stably maintain their improvement many years after implantation.

The present study is not without inherent limitations. This is a single-institution study, where all of the subjects underwent standardized surgical treatment with no observation cohort for comparison. Future, prospective studies including an observation cohort are warranted to validate the current findings. Although this is the largest study to date regarding outcomes in SSD and CI, the preoperative etiology groups vary in their makeup, demographics, and follow-up time. The variability in follow-up time has the potential to introduce bias, which is an inherent limitation. The tumor group's small size limits statistical analysis to provide conclusive results. Despite these inherent limitations, the data support the benefits of CI in rehabilitation of subjects with SSD consistent with previous studies.

CONCLUSION

The current study provides the largest analysis of subjects treated with a CI for SSD with data collected longitudinally over an extended period of time. We found that subjects functionally improve with respect to their speech perception and binaural benefit with improved head shadow effect, and they gradually improve with sound localization after multiple years of auditory rehabilitation. These findings further support the benefit of a CI in subjects with a deafened ear.

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