Human Otopathology of Cochlear Implant Drill-out Procedures

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Abstract

Objective. Human otopathology following drill-out procedures for cochlear implantation (CI) in cases with labyrinthitis ossificans (LO) has not been previously described. This study uses the high sensitivity of histopathology to (1) evaluate surgical drill-out technique with associated intracochlear findings and (2) quantify spiral ganglion neuron populations in a series of patients with LO who underwent CI.

Study Design. Retrospective otopathology study.

Setting. Otopathology laboratory.

Subjects and Methods. Temporal bone (TB) specimens from cases with evidence of preoperative intracochlear fibroossification that required a drill-out procedure for CI electrode array insertion were included. All cases were histopathologically evaluated and 3-dimensional reconstructions of the cochleae were performed to interpret drilling paths and electrode trajectories.

Results. Five TB specimens were identified, of which 4 underwent drill-out of the basal turn of the cochlea and 1 underwent a radical cochlear drill-out. In multiple TBs, drilling was imprecise with resultant damage to essential structures. Two TBs showed injury to the modiolus, which was associated with substantially decreased or even absent neuronal populations within these areas. In addition, 2 cases with inadequate drill-out or extensive LO of the basal turn resulted in extracochlear placement of electrode arrays into the vestibule due to persistent obstruction within the basal turn.

Conclusion. Otopathology highlights the challenges of drill-out procedures in cases of LO. Imprecise drilling paths, due to distortion of normal cochlear anatomy, risk injury to the modiolus and adjacent neurons as well as extracochlear placement of electrode arrays, both of which may contribute to poorer hearing outcomes.

Keywords

cochlear implantation, labyrinthitis ossificans, otopathology

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the extent of LO may be associated with lower spiral ganglion neuron (SGN) populations. Thus, it remains unclear if poor hearing outcomes are related to preoperative characteristics of patients with LO or due to the challenges of drill-out procedures and appropriate electrode array placement secondary to extensive ossification.

While the histopathology of LO in cases of bacterial labyrinthitis, otosclerosis, and autoimmune inner ear disease has been described previously, little is known about cochlear histology following drill-out procedures for placement of CI. In fact, no prior study has rigorously assessed intracochlear histopathologic changes occurring as the result of drill-out procedures for CI. The aim of this study is to (1) evaluate the consequences of the CI surgical technique, including intracochlear findings, and (2) quantify postsurgical SGN populations in a series of patients with LO who underwent CI requiring a drill-out for electrode array placement.

Methods

Specimens

The National Temporal Bone Hearing and Balance Pathology Resource Registry was used to identify all cases of patients with a history of cochlear implantation. Included cases had (1) evidence of intracochlear fibro-ossification on preoperative imaging (ie, intracochlear opacity) or at the time of CI surgery that required a cochlear drill-out procedure and (2) a preoperative diagnosis known to result in LO, including meningitic labyrinthitis (ML), autoimmune inner ear disease (AIED), temporal bone fracture, and otosclerosis. Exclusion criteria included (1) CI without a cochlear drill-out procedure and (2) severe postmortem autolysis that prevented SGN quantification (Figure 1). This study was exempted by the Massachusetts Eye and Ear Infirmary Institutional Review Board.

Histological Techniques

TB specimens were fixed in 10% buffered formalin and processed by methods previously described. TBs were embedded in araldite with the electrode in situ or in celloidin with the electrode removed and sectioned in the horizontal plane (20-μm thickness). Sections were then stained with osmium, toluidine blue, or hematoxylin and eosin and mounted on glass slides for analysis.

Two-Dimensional Reconstruction of the Cochlea

Every 10th section of TB was studied under light microscopy, and the cochlear duct and Rosenthal’s canal were reconstructed by 2-dimensional (2-D) methods, previously described. The degree of LO was classified into 3 categories of severity based on the percentage of the cochlear lumen affected: mild (<25% of lumen), moderate (25%-75% of lumen), and extensive LO (>75% of lumen). SGNs were counted within each segment of Rosenthal’s canal and normalized by age-matched populations (controls) from individuals with useful hearing. The degree of SGN loss was classified into 4 categories: mild (≥80% control), moderate (50%-79% control), severe (20%-49% control), and profound (<20% control).

Three-Dimensional Reconstruction of the Cochlea

Three-dimensional (3-D) reconstruction of the cochlea was performed by obtaining digital images of every 10th slide using a light microscope with a high-resolution camera (Olympus BX51, Olympus DP71; Olympus, Tokyo, Japan). Images (range of 30-40 for each specimen) were aligned, converted to grayscale, and segmented using Amira software (version 6.0.0; FEI, Hillsboro, Oregon). All images were analyzed and each 2-D image was segmented into the following structures: otic capsule, drill-out path, cochlear lumen, round window membrane (RW), carotid canal, and electrode. This allowed for 3-D interpretation of drilling paths and electrode trajectories. To determine safe drilling practices and to provide an additional surgical landmark in basal turn drill-out, a mean maximum safe drilling depth from the RW was determined. This measure was made from the anterior-inferior edge of the RW to the anterior aspect of the otic capsule of the lower basal turn.

Results

Patient Demographics and Etiology of Hearing Loss

Five specimens were identified that underwent a drill-out procedure for CI during life and showed evidence of intracochlear fibro-ossification on preoperative imaging or at the time of surgery (Table 1). This cohort included 2 left and 3 right ears, with cases 3 and 4 from a single patient who underwent sequential bilateral CI. Four cases were from males. The etiology of hearing loss (HL) was meningitic labyrinthitis in 2 specimens and Cogan’s syndrome (CS) in 1 specimen. Two additional specimens had a clinical history...
consistent with CS but were not definitively diagnosed during life. Prior to CI, all patients had documented severe to profound bilateral sensorineural hearing loss. The median duration of HL prior to CI and duration of CI-use were 10.3 (range, 0.3-24.7) years and 15.1 (range, 4.1-23.3) years, respectively.

**Cases of Drill-out with Scala Vestibuli Insertion of Electrode Arrays**

Two cases required drill-out of the basal turn for CI, with intracochlear placement of the electrode array. In case 1, preoperative imaging showed evidence of intracochlear obstruction. Preoperative imaging was also performed for case 2, but available reports did not indicate whether there was evidence of intracochlear obstruction. In both cases, an ossified cochlea was encountered, but the lumen of the basal turn was successfully identified. Further drill-out of the basal turn revealed a patent distal lumen that allowed for complete insertion of the CI electrode. Postoperative word testing was never performed in case 1 due to cognitive delay and death at a young age, but clinical notes indicate the patient wore his device during all waking hours and consistently responded to his name. Case 2 had 40% NU-6 word scores at 1 and 5 years post-CI.

Histological analysis of case 1 showed moderate LO of the ST and SV within the lower basal turn of the cochlea and severe LO within the distal cochlea (Figure 2A,B). Case 2 had mild to moderate LO of the ST and SV within the lower basal turn, with a relatively patent lumen within the distal cochlea (Figure 2C,D). Both cases showed drilling extending over the entire inferior aspect of the basal turn, with an electrode path entering into the SV of the basal turn. SGN counts for case 1 showed moderate to severe loss within segments I to II of Rosenthal’s canal (34%-61% of control) and profound loss within the heavily ossified segments III to IV (0%-2% of control). SGN counts for case 2 showed moderate to severe loss throughout all segments of Rosenthal’s canal (44%-73% of control) (Table 2).

**Cases of Drill-out with Extracochlear Placement of Electrode Arrays**

Two cases, from a single patient, required extensive drill-out of the round window niche for CI, but resulted in extracochlear placement of electrode arrays, as identified on histological analysis. Cases 3 and 4 had preoperative imaging that was reported as negative for intracochlear ossification. Both cases showed extensive drilling of the basal turn of the cochlea in the area of the round window. Case 3 was a reimplant case; fibrous tissue was noted in the area of the round window at reimplantation, and incomplete insertion of only 15 of 22 electrodes of a Nucleus (Cochlear Ltd, Sydney, Australia) device was achieved due to resistance and kinking of the electrode array. Clinical records for case 4 note that extensive drilling of the round window area was required for implantation. Insertion of only 4 of 6 total

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**Table 1. Patient and CI Characteristics.**

<table>
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<tr>
<th>Operative Report</th>
<th>Case No.</th>
<th>Age at Death, y</th>
<th>Cause of HL</th>
<th>Age at Onset, y</th>
<th>Years of HL</th>
<th>Side CI</th>
<th>Age at CI, y</th>
<th>HL Prior to CI, y</th>
<th>Duration of CI-use, y</th>
<th>HL Prior to CI, y</th>
<th>CI Use, y</th>
<th>Hearing Results</th>
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<tr>
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<td>1</td>
<td>6 ML</td>
<td>L Nucleus</td>
<td>19</td>
<td>1</td>
<td>R</td>
<td>4.4</td>
<td>0.3</td>
<td>10.3</td>
<td>0.3-24.7</td>
<td>CT</td>
<td>Basal drill-out SV, full</td>
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<tr>
<td></td>
<td>2</td>
<td>47 CS</td>
<td>R Ineraid</td>
<td>27.5</td>
<td>2</td>
<td>L</td>
<td>8.9</td>
<td>0.3</td>
<td>10.5</td>
<td>0.3-24.7</td>
<td>CT</td>
<td>Basal drill-out SV, complete</td>
</tr>
<tr>
<td></td>
<td>3</td>
<td>89 CS</td>
<td>R Ineraid</td>
<td>28.0</td>
<td>1</td>
<td>L</td>
<td>8.9</td>
<td>0.3</td>
<td>10.3</td>
<td>0.3-24.7</td>
<td>CT</td>
<td>Basal drill-out Incomplete (15 of 22 EL)</td>
</tr>
<tr>
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<td>4</td>
<td>89 CS</td>
<td>L Ineraid</td>
<td>41</td>
<td>0</td>
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<td>0.3</td>
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<td>Basal drill-out Incomplete (3 of 6 EL)</td>
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<td>10.3</td>
<td>0.3-24.7</td>
<td>CT</td>
<td>Radical cochleotomy</td>
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**Abbreviations:** CI, cochlear implantation; CS, Cogan’s syndrome; CSF, cerebrospinal fluid; CT, computed tomography; EL, electrodes; HL, hearing loss; L, left; ML, meningitic labyrinthitis; NU-6W, Northwestern University Auditory Test Number Six word score; R, right; SD, speech discrimination; SV, scala vestibuli.

*Initial Symbion-Ineraid electrode explanted during reimplantation with a Nucleus 22 electrode.*
Electrodes of a Symbion Ineraid (Symbion Inc, Salt Lake City, Utah) was achieved. While limited details on the operative procedures are available, patient positioning in these cases appears to have been suboptimal due to cervical arthritis, which limited the surgeon’s view through the facial recess. All surgeries were performed by experienced surgeons, but their specific familiarity with cochlear drill-out is unknown. Postoperative imaging reports indicated “intracochlear placement” of the electrode in case 3 and placement of the electrode into a “drilled-out groove in the basal turn” for case 4. Neither CI provided any speech discrimination.

On histological analysis, case 3 showed severe LO of the ST within the lower basal turn with an otherwise patent cochlear lumen (Figure 3A,B). The specimen showed drilling over the hook of the cochlea with direct violation of segment I of Rosenthal’s canal. Three-dimensional reconstruction confirmed a drill path extending over the hook and vestibule and angled medially toward the modiolus (Figure 3C,D). In addition, SGN counts showed profound loss within segment I (19% of control) and mild to moderate loss within segments II to IV (54%-87% of control) (Table 2). While an electrode track is observed entering into the cochlear hook, both 2-D and 3-D analyses show the electrode deflecting off an ossified basal turn, with resultant extracochlear placement into the vestibule (Figure 3).

Histological analysis of case 4 showed severe LO of the entire cochlear lumen. Drilling was observed over the basal turn with extension into the vestibule, without violation of Rosenthal’s canal (Figure 4A,B). SGN counts showed severe to profound loss throughout the entire cochlea (23%-40% control) (Table 2). An electrode track is observed with a trajectory toward the basal turn of the cochlea but appears to deflect off an ossified basal turn, turning posterosuperiorly into the vestibule (Figure 4).

Case of Radical Cochlear Drill-out
Case 5 had no record of preoperative imaging, but after encountering intracochlear ossification, the lumen of the
basal turn was identified and a radical cochleotomy was performed. The electrode was then successfully placed into the remnant middle turn of the cochlea and packed in place with temporalis fascia. A cerebrospinal fluid (CSF) leak and bleeding from periarterial tissue surrounding the carotid artery were noted at the time of surgery, but there was no mention of neurologic complications from arterial injury. This patient had poor performance with /C20^16% NU-6 word scores, which may, in part, have been hindered by recurrent meningitis. He died 15 years later of what was thought to be recurrent bacterial meningitis.

Histological analysis showed severe LO of the entire cochlear lumen. Extensive drilling was observed over the basal turn and into the lower middle and apical turns, with direct violation of segments III and IV of Rosenthal’s canal and anterior extension through the bony carotid canal (Figure 5A,B). SGN counts showed profound loss in segments I to II, although there was evidence of some remaining neurons (4%-12% of control) (Figure 5C); no SGNs were observed in the remaining portions of segments III to IV (Table 2). Three-dimensional reconstruction confirmed a drill path over the inferior basal turn, with superior extension into the middle and apical turns and medial extension into the modiolus (Figure 5D). An electrode track was observed within the remnant middle turn of the drilled-out cochlea.
Maximum Safe Drilling Depth from Round Window Membrane to Anterior Basal Turn of Otic Capsule

Within this cohort, the average distance between the RW and anterior aspect of the otic capsule of the basal turn was 7.3 mm (range, 6.8-7.9 mm) (Figure 6).

Discussion

In this otopathologic study, we evaluate cochlear histopathology and audiometric outcomes in cases of labyrinthitis ossificans and CI requiring drill-out for electrode array placement. As the first histologic study of its kind, we find that drilling was frequently imprecise with resultant damage to essential structures. Injury to the modiolus was identified in 2 specimens and was associated with substantially decreased or even absent neuronal populations within these areas. In addition, inadequate drill-out with extensive ossification of the basal turn was found to result in extracochlear placement of electrode arrays into the vestibule due to persistent obstruction of the cochlear lumen.

Two cases of drill-out procedures with intracochlear electrode placement were identified. In both, ossification of the lower basal turn was overcome by identification of the lumen of the basal turn, drill-out procedure, and subsequent electrode insertion into the scala vestibuli. No gross complications were noted on histological analysis, and reported hearing outcomes were moderate, with 40% word recognition score (WRS) in 1 patient. Prior clinical studies have shown similar hearing results with scala vestibuli insertion of electrodes, although published studies may be limited by the inability to identify scalar location of the electrode with intraoperative plain film. Together, these findings suggest that scala vestibuli insertion following drill-out in the correct trajectory may be an effective electrode placement option for CI in select cases of bony obliteration of the scala tympani.

Two cases with varying degrees of ossification resulted in extracochlear placement of the electrode following drill-out of the round window area. In case 3, an inappropriate trajectory of drill-out and attempted insertion against resistance appears to have resulted in deflection of the electrode into the vestibule. Similarly, case 4 showed inadequate drill-out of the basal turn, resulting in deflection of the electrode into the vestibule. While intracochlear ossification may not have been expected, these cases highlight the need to adapt surgical procedures if obstruction or resistance to insertion is noted intraoperatively. Identification of the lumen of the basal turn is essential and an adequate drill-out to a patent lumen must be performed to ensure proper electrode placement. New bone within the cochlear lumen may be differentiated from the otic capsule intraoperatively as it is often slightly malleable before it is fully ossified and
In cases where a patent cochlear lumen cannot be identified, a circumodiolar drill-out is necessary and should be coupled with blind sac closure of the external auditory canal for adequate cochlear visualization and secondary prevention of spinal fluid leak.10

One case with extensive ossification requiring a radical cochlear drill-out demonstrated multiple complications. Drill-out of the middle turn extended too far medially into the modiolus and superiorly into the apical turn, with resultant damage to remaining SGNs. The operative report also suggests a CSF leak, likely through the damaged modiolus. Damage to the carotid canal also occurred when drilling extended too far anteriorly. We found that this can be prevented by limiting drill-out of the basal turn to approximately 7 mm anterior to the RW membrane.

Distortion of normal cochlear anatomy in cases of severe labyrinthitis ossificans highlights the need for intraoperative confirmation of electrode placement.21 Proper patient positioning and imaging in the Stenvers projection can accurately confirm intracochlear placement of an electrode array with a single exposure.21 Intraoperative radiography can provide an opportunity to remove and reinsert the electrode at the time of initial placement. With particularly challenging cases where surface anatomy alone is inadequate for identification of the cochlear scalae, high-resolution intraoperative image guidance may be warranted.22 If auditory performance is poor following activation, postoperative high-resolution computed tomography is indicated, and a revision procedure by an experienced CI surgeon should be planned if the electrode position is suboptimal.

Postoperative spiral ganglion neurons were decreased when compared to age-matched controls for all patients in this study, which aligns with prior studies showing a negative association between neuronal populations and intracochlear ossification.11,12 However, surviving neurons occurred in segments both affected and unaffected by labyrinthitis ossificans; therefore, even cases of extensive ossification may maintain sufficient neuronal populations for CI function. Given the low SGN populations in LO, careful technique for cochlear drill-out is essential. In fact, 2 cases with drilling misdirected into Rosenthal’s canal showed profound loss of SGNs within these areas. In one, drilling was angled too far medially into the hook, and in another, drilling of the middle turn extended too far medially into the modiolus and superiorly into the apical turn.

This study is limited by its retrospective nature, small sample size, and available clinical data. Electrode model may affect results, and limited available cases prevented comparison of a single type of electrode. In addition, 1 patient was a reimplantation case, which may affect intralabyrinthine observations and hearing outcomes. In cases without available preoperative imaging, pre-CI intracochlear ossification was determined from operative reports, which may vary in accuracy. In addition, labyrinthitis ossificans is a progressive disorder, and some degree of progression of ossification may have occurred following surgery or as the direct result of CI.

This is the first study to demonstrate histopathologic findings following drill-out for cochlear implantation in patients with labyrinthine ossification. Novel otopathologic findings illustrate that SGNs persist, despite the presence of extensive ossification. Furthermore, destruction of normal anatomy, including neuronal loss and suboptimal electrode placement following cochlear drill-out, may explain variable audiometric outcomes. Findings highlight the importance of identification of the lumen of the basal turn and precise insertion technique as misaligned array position may further degrade outcomes in an already “fragile” cochlea. Intraoperative image guidance may be one technique to obtain appropriate drill-out trajectory in the future, but this technology requires further study in cases of labyrinthitis ossificans.

Conclusion

In this human otopathologic study, we find that drill-out paths in cases of labyrinthitis ossificans may be imprecise due to distortion of normal cochlear anatomy. Multiple cases highlight the risk of direct injury to limited neuronal populations and show that incomplete drill-out or severe ossification of the cochlear basal turn may lead to extracochlear placement of electrode arrays, both of which may contribute to poorer hearing outcomes.

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Author Contributions

Danielle R. Trakimas, acquisition of data, analysis and interpretation of data, final approval of version to be published, and agreement to be accountable for all aspects of work; Reuven Ishai, interpretation of data and revising content, final approval of version to be published, and agreement to be accountable for all aspects of work; Elliott D. Kozin, interpretation of data and revising content, final approval of version to be published, and agreement to be accountable for all aspects of work; Joseph B. Nadol Jr., interpretation of data and revising content, final approval of version to be published, and agreement to be accountable for all aspects of work; Aaron K. Remenschneider, acquisition of data, analysis and interpretation of data, drafting and revising content, final approval of version to be published, and agreement to be accountable for all aspects of work

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