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Otopathology of Unilateral Cochlear Implantation in Patients with Bilateral Temporal Bone Fracture

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Abstract

Objective: Audiometric outcomes following cochlear implantation (CI) in patients with temporal bone fracture (TBF) are variable and the pathophysiology is not well understood. Herein, we describe otopathologic findings in two such cases to provide insight into pathophysiology and outcomes.

Patients: Two patients had a history of head trauma resulting in bilateral TBF and sudden, profound, sensorineural hearing loss.

Intervention: Both cases underwent unilateral CI following head trauma.

Main Outcome Measures: Both TBs from each patient were harvested postmortem and histologically analyzed to determine fracture lines, intralabyrinthine fibroosseous changes, and spiral ganglion neuron (SGN) counts. Clinical histories and audiometric outcomes were also reviewed.

Results: In both cases, the implanted TB showed extensive intracochlear ossification, which was greater when fracture involved the cochlea. In contrast, the non-implanted cochleae were patent with small osseous deposits at the round window membrane. Interaural SGN counts were lower on the implanted side and hearing results were moderate to poor.

Conclusions: Otopathologic analyses of patients with a CI after bilateral TBF are rare. Based on otopathology of two patients with bilateral TBF and subsequent CI, implanted TBs show extensive intracochlear fibroosseous changes greater than the non-implanted side. Findings implications for auditory rehabilitation after TBF.

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Introduction

The otologic sequelae of temporal bone fracture (TBF) are heterogeneous and depend on the location and trajectory of the fracture line, with complications including hearing loss, facial nerve paralysis, and meningitis.¹ In terms of hearing loss, fractures may result in sensorineural hearing loss (SNHL) due to disruption of the sensory neuroepithelium.² Acute intracochlear hemorrhage and delayed labyrinthitis ossificans (LO), may also lead to profound SNHL.^{3–5}

Cochlear implantation (CI) or auditory brainstem implantation (ABI) remain the only options for auditory rehabilitation for patients with profound SNHL following TBF. While CI is preferred in most cases, outcomes following TBF are variable and the pathophysiologic basis for this variability is poorly understood. Limited reports of small patient cohorts indicate outcomes ranging from poor to excellent word discrimination, with variability of outcomes most commonly attributed to duration of hearing loss.^{4,6,7} Anatomic changes to the cochlea and labyrinth occur following TBF and may also affect outcomes following CI. Chronic changes following fractures, such as LO, may complicate electrode insertion⁸ and require careful pre-operative planning.^{4,6,7}

Histopathology of CI after TBF has not been rigorously described. An understanding of intracochlear changes in such cases may provide important insight into clinical management and prognosis in head trauma patients.⁹ To better understand pathophysiology and outcomes, we evaluated otopathologic findings in two patients who underwent CI after TBF.

Materials and Methods

Specimens

TB specimens from the Otopathology Laboratory (Boston, MA) having undergone CI were evaluated. Inclusion criteria consisted of cases with 1) both TBs available and 2) history of bilateral TBF. Exclusion criteria consisted of a history of meningitic labyrinthitis.

Histological Techniques

TBs were fixed in 10% buffered formalin and decalcified in ethylenediaminetetraacetic acid. The TBs were dehydrated in alcohol and embedded in araldite with the electrode *in-situ*¹⁰ or in celloidin with the electrode removed, for sectioning. Sections were cut in a horizontal plane (20µm thickness), stained with osmium, toluidine blue or hematoxylin and eosin and then mounted on glass slides.¹¹ Every tenth section of the TB was studied and the cochlear duct and Rosenthal's canal were reconstructed by conventional 2-D methods.¹¹

Histologic and Clinical Outcomes Assessment

Segmental numbers of SGN were counted and normalized by age-matched controls.^{11,12} The difference in normalized SGN was calculated between the implanted and non-implanted ear within each patient:¹³ $SGN_{\text{implanted ear}}(\%) - SGN_{\text{non-implanted ear}}(\%) \times 100\%$. Pre-operative and post-operative audiometry and speech discrimination were analyzed.

Results

Brief Case Summary and Trauma History

Two patients met inclusion and exclusion criteria. Both cases had a history of head trauma resulting in bilateral TBF and bilateral sudden, profound SNHL. Each patient underwent subsequent CI in one ear (Table 1). Prior to CI, both patients showed evidence of vestibular hypofunction on caloric testing. Rotatory chair testing was not described to confirm the vestibular weakness.

Case 1: The right, implanted, TB shows longitudinal and transverse otic capsule fractures. The longitudinal component involves the apical and middle turns of the cochlea. Multiple transverse fractures involve the fallopian canal (FC), internal auditory canal (IAC), stapes footplate (SFP), promontory and posterior semicircular canal (PSCC) (Figures 1A,B). The left, non-implanted, TB shows a transverse otic capsule fracture similar to that on the contralateral side (Figures 1C,D).

Case 2: The left, implanted, TB is incomplete with part of the vestibular labyrinth lost during TB harvesting. A transverse otic capsule fracture involves the promontory and PSCC (Figure 2A) with an adjacent thin layer of fibrous tissue indicating healing fracture lines (inset). The right, non-implanted, TB shows a transverse otic capsule fracture involving the basal turn of the cochlea (Figure 2B), promontory and PSCC.

Cochlear Implant Insertion

Both cases had full insertion of the electrode array: 1) Case 1 was implanted with a Nucleus 22 device 6-months post-trauma and died 2 years later from unknown causes; 2) Case 2 was implanted with a single channel electrode by a round window approach 6 years post-trauma and underwent ipsilateral reimplantation due to poor performance with a Nucleus 22, 5 years later. The patient died 10 years later from lung cancer. As further described below, both electrodes were inserted into the scala vestibuli (SV). Available medical records do not provide insight into whether this was the intended surgical approach.

Audiometric Outcomes

Prior to CI, both patients had documented bilateral profound SNHL with no speech discrimination. Last available audiometry for Case 1 documented 30% on monosyllabic word testing at 2-years post-CI, while Cases 2 had 4% CID sentences 2 years after reimplantation.

Otopathology of Implanted Temporal Bones

Cases show drilling of the otic capsule over the SV and the CI electrode tract enters the SV surrounded by a thin layer of fibrosis (Figures 3A,B). In Case 1, the electrode remains in the SV throughout the basal turn, and in Case 2, the electrode translocates to the scala tympani (ST) within the ascending basal turn. Neither case showed evidence of fracture of the osseous spiral lamina (OSL) or modiolus. On the implanted side, both TBs showed substantial intracochlear ossification. Case 1 had partial ossification of the ST and SV, which extended 12 mm beyond the electrode tip, into the apical turn of the cochlea. Case 2 showed complete ossification of the ST and SV within the proximal basal turn with partial fibroossification of the distal basal turn. In both TBs, SGN counts were decreased to 30-75% of age-matched controls within Segments I-II of Rosenthal's canal. In Segments III-IV, SGN counts were generally higher, ranging from 50-85% of age-matched controls (Table 1).

In both cases, the lateral semicircular canal (LSCC) showed either partial (Case 1) or extensive (Case 2) fibroossification. The remainder of the vestibular labyrinth remained patent in Case 1, with partial fibroossification of the vestibule and semicircular canals in Case 2. Case 2 had evidence of a traumatic neuroma that was described previously (Figure 2B).¹⁴

Otopathology of Non-Implanted Temporal Bones

Non-implanted TBs showed different findings within the cochlea compared to the implanted side: both cochleae were patent with a small osseous deposit at the round window membrane (Figures 3C,D). Similar to the contralateral side, neither case showed evidence of fracture of the OSL or modiolus. In Case 2, SGN counts were higher than the implanted side throughout the cochlea, except within Segment III, where fracture involved the cochlea (Table 1). Case 1 showed severe autolysis that prevented SGN quantification. The vestibular labyrinth, however, showed comparable fibroosseous changes between sides of each case.

Discussion

Otopathologic analysis of patients that sustained bilateral TBF without meningitis and subsequent CI placement is rare, and evaluation is critical to understand the pathophysiology, prognosis and surgical approach for auditory rehabilitation. In two cases of bilateral TBF and subsequent CI, specimens showed ossification of the cochlea on the implanted side, which was greater than that observed on the non-implanted side. SGN counts were decreased on the implanted side, and Case 2 showed relative interaural losses on the implanted side in all segments apart from that adjacent to fracture involving the cochlea on the non-implanted side. While the non-implanted side showed fibroossification of the vestibular labyrinth, the cochlea remained patent, even with fracture involving the cochlea. It is notable that the surgeon implanted the side with more extensive fractures. It is not known whether the surgeon was aware that one side was more severely fractured than the other prior to selection of the ear for CI.

Ossification of the vestibular labyrinth was observed in all TBs and was most prominent within the LSCC. While one may assume this is due to direct disruption of the vestibule and

semicircular canals, this response did not occur with fracture of the cochlea in the absence of CI. Additionally, intravestibular osseous changes were similar between ears of each case, suggesting this effect is independent of CI.

Few previous studies have discussed otopathologic findings of CI after TBF.^{9,15} Lee et al. briefly described three cases in the setting of electrode insertion.¹⁵ Insertion was complete in all three cases of TBF despite a transverse fracture through the basal, middle and apical turn in one case, and new bone formation was present in the ST and SV in the ascending limb of the basal turn in 2 cases.¹⁵ The report, however, does not provide information regarding presence of meningitis, exact location of osseous changes, SGN counts or audiometric outcomes nor does it contain otopathology figures.

Previous otopathologic studies of CI patients without TBF show that mild intracochlear ossification can occur after CI,¹⁶ usually adjacent to the electrode with greater ossification observed in cases of traumatic or improper electrode insertion.¹⁶ In both cases, we observe substantial intracochlear ossification on the implanted side. When fracture involved the cochlear labyrinth on the implanted side, ossification extended far beyond the electrode tract, which prior studies show is not typically observed following CI. While TBF itself may lead to LO⁸, we did not observe these changes within the cochlea on the contralateral, non-implanted side even when fracture involved the cochlea. Trauma to the OSL could also initiate new bone formation and eventually affect the hearing performance of the CI users.¹⁷ However, we did not see any evidence of damage to the OSL, neither by fracture or electrode-induced trauma.

The retrospective case series has several limitations. One patient was implanted twice on the same side; however, the initial CI involved round window insertion of a short electrode, which should cause minimal acute and chronic intracochlear changes.¹⁸ Additionally, it is possible the cochlear ossification following CI would have been comparable without the fracture. It is important to highlight that fracture patterns differed between sides and cases, which could theoretically result in differential shear forces and injury to neuronal structures. The implanted side of Case 1, in particular, showed an extensive fracture involving the cochlea, which may, in part, contribute to greater intracochlear changes and SGN loss on this side. Despite these limitations, this is the first report of long-term otopathologic changes following CI in patients with bilateral TBF.

Taken together, these cases suggest that TBF is not always associated with intracochlear LO and that full insertion of a CI, even years later, is possible. We hypothesize that TB fracture that involves the cochlear labyrinth shows substantial intracochlear ossification that extends beyond the electrode tract and may represent a “fragile” ear that is prone to more extensive fibroosseous changes following CI, however, this hypothesis needs to be further investigated. Future studies are needed to understand how TBF may impact cochlear physiology to result in fibroosseous deposition.

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Figure 1: Case 1 TB specimens.

A&B: Horizontal section of right TB stained with toluidine blue showing a longitudinal fracture (Fx) involving the apical and middle turns of the cochlea and a transverse fracture involving the stapes footplate (SFP). There is an electrode tract (red *) in the scala vestibuli (SV) of the basal turn with dense new intracochlear bone (orange +) and fibrosis (>). C&D: Horizontal section of left TB stained with osmium showing a transverse fracture involving the SFP and vestibule (Vest) with new bone (blue +) and fibrous tissue (>) formation in the lateral semicircular canal (LSCC) and Vest. ME: middle ear, IAC: internal auditory canal, FN: facial nerve.

Figure 2: Case 2 TB specimens.

A: Horizontal section of left TB stained with hematoxylin and eosin showing a transverse fracture (Fx) involving the promontory. There is an electrode tract (red *) in the basal turn with dense new bone (orange +) and fibrous tissue (>). The inset shows evidence of fibrous healing adjacent to the fracture line. B: Horizontal section of right TB stained with hematoxylin and eosin showing a transverse fracture involving the spiral ligament (SL, o) of the basilar turn and internal auditory canal (IAC). There is new bone in the vestibule (Vest) and common crus (CC). FN: facial nerve.

Figure 3: Horizontal sections through basal turn of TB specimens.

A: Right, implanted TB from Case 1. The electrode (red *) enters the cochlea (COCH site) into the scala vestibuli (SV). There is dense new bone (blue +) in the cochlea and a fracture (Fx) involving the posterior semicircular canal (PSCC). B: Left, implanted TB from Case 2 shows the electrode entering the SV. Dense new bone fills the cochlea and PSCC. A fracture involves the PSCC and the inset shows evidence of fibrous healing adjacent to the fracture. C: Left, non-implanted TB from Case 1. A fracture involves the promontory and PSCC and a small osseous deposit is seen at the round window membrane (RWM). D: Right, non-implanted TB from Case 2. A fracture involves the PSCC with complete ossification of the PSCC and a small osseous deposit at the RWM.

Table 1:

Patient demographics, surgical history, and otopathologic findings of both cases. Fracture type: transverse (Tr), longitudinal (Lo), or complex (Co). Healing of fracture with: fibrosis (F) and/or ossification (O). Ossification is noted as none, minimal (min), partial (part), or extensive (ext) within the scala tympani (ST) and scala vestibuli (SV) throughout Segments I-IV of Rosenthal's Canal and within the superior/lateral/posterior semicircular canal (SSCC/LSSCC/PSSCC) and vestibule (Vest). Spiral ganglion neuron (SGN) counts are presented as % of age-matched controls and the difference between ears (= implanted – non-implanted).

Demographics				Pre-op PTA (dB)		Surgical information			TB fracture		Intralabyrinthine osseous changes		SGN (%)				Post-op hearing							
Pt	Sex	Age at trauma (yr)	Cause of trauma	Age at death (yr)	R	L	Age at CI (yr)	Duration of deafness before CI (yr)	Side	CI	Extent of insertion	Type	Healing	Cochlear	Vestibular	Seg I	Seg II	Seg III	Seg IV	Total	Timing after CI (mo)	Test	Score	
1	M	64	Syncope	67	NR	NR	65	0.5	R	Nucleus 22	Full	Co	F	ST: part in Seg I-III SV: part Seg I-IV	LSCC: part SSCC, PSCC, Vest: none		43	31	50	48	42	24	monosyll words	30%
2	M	47	Fall	67	NR	NR	53 58	6	L	House single channel Nucleus 22	n/m	Tr	F	ST: part Seg I-II SV: part Seg I-II	LSCC: ext SSCC, PSCC, Vest: part		43	75	81	86	75	14	CID sent	4%
									R	none	n/a	Tr	F & O	ST: RWM SV: none	LSCC, PSCC: ext SSCC, Vest: part		104	82	71	87	83	-	-	-
																	-61	-7	10	-1	-8		-	