




## OTOPATHOLOGY REPORT

# Otosclerosis and the evolution of stapes surgery: A historical and otopathological study

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**Abstract**

**Objective:** To explore the historical evolution of surgical techniques for otosclerosis treatment, viewed through the lens of human temporal bone pathology to aid in understanding the disease and refining surgical interventions.

**Data sources:** A review of historical literature on otosclerosis, surgical techniques, and otopathological findings was conducted. Eight temporal bone specimens from the Paparella Otopathology & Pathogenesis Laboratory, University of Minnesota, and one from the University of California, Los Angeles, were analyzed.

**Review methods:** We selected two temporal bones from donors who underwent four different types of surgical procedures for otosclerosis: stapes mobilization, fenestration, stapedectomy, and stapedotomy. One successful and one complication case was selected for each procedure. Histopathological analysis was performed to assess the outcomes and complications associated with each technique.

**Results:** The study chronicles the progression of otosclerosis surgery from the stapes mobilization to modern stapedectomy and stapedotomy techniques. Initial procedures, like stapes mobilization and fenestration, yielded limited and temporary results with significant complications. The introduction of stapedectomy marked a significant improvement, with better long-term outcomes. Histopathological analysis revealed insights into the causes of surgical failures and complications.

**Conclusion:** Otosclerosis surgery has evolved significantly, driven by advances in otopathology and surgical technology. While earlier techniques offered limited success, modern procedures like stapedectomy and stapedotomy provide improved outcomes and fewer complications. Ongoing research promises further advancements in the field, improving patient care and surgical efficacy.

**Level of evidence:** NA.

**KEYWORDS**

inner ear, otology, otopathology, otosclerosis, stapes surgery, temporal bone pathology

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## 1 | INTRODUCTION

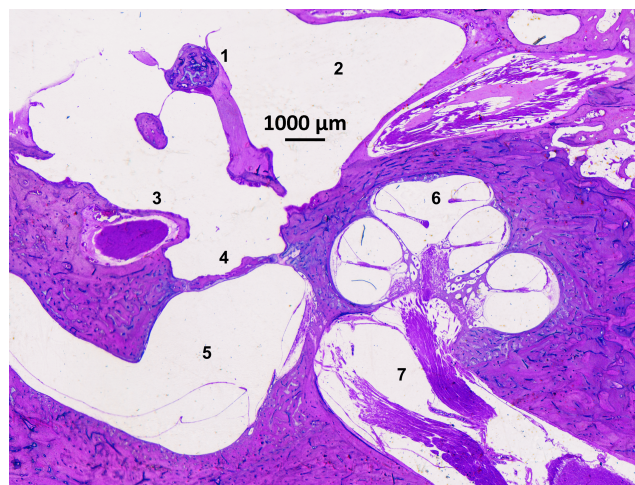
Otosclerosis was one of the first diseases affecting hearing that was identified through anatomic and histopathological study. The first description of otosclerosis occurred during a postmortem examination conducted by Dr. Antonio Valsalva in 1735. He observed ankylosis of the stapes around the oval window and referred to this fixation as “dry catarrh of the middle ear.”<sup>1,2</sup> Subsequently, several authors in the 19th century reported similar pathology in their patients. However, it was not until 1860 that Joseph Toynbee recognized ankylosis of the stapes as a common cause of deafness, finding stapes fixation in 35 temporal bone dissections.<sup>3</sup>

Anton von Troeltsch, in 1881, was the first author to use the term “sclerosis” to describe these changes that were previously attributed to chronic interstitial middle ear inflammatory mucosal changes. The underlying pathology was later identified by Adam Politzer in 1894, when he studied 16 post mortem patients with stapes fixation and realized that the fixation was not caused by secondary ankylosis, but actually due to a primary disease of the bony labyrinthine capsule.<sup>4</sup> Thus, he added the “oto” to the name, creating the disease name known today—otosclerosis—which is Greek for “hard ear condition.”<sup>5</sup>

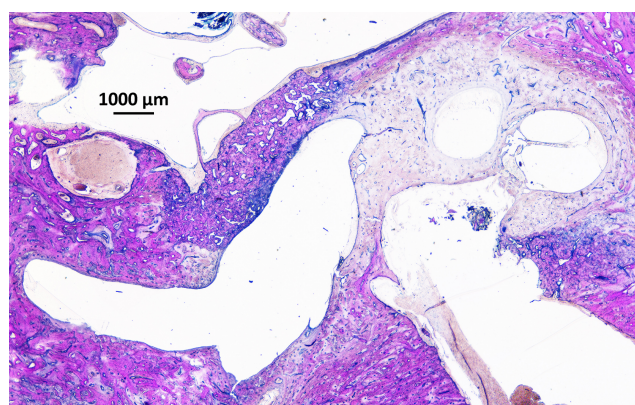
Following Politzer's discovery, numerous findings contributed to clinicians' enhanced understanding of otosclerosis. Associations between otosclerosis and sensorineural hearing loss (SNHL) were also established, suggesting involvement of the cochlear endosteum.<sup>6</sup> Siebmann introduced the term “otospongiosis” to better describe active stages of otosclerosis. This recognition led to the understanding that the term otosclerosis alone did not adequately capture the disease process. Although more precise terms have been coined, otosclerosis has remained the commonly used name for the condition, despite its incomplete description of the changes that occur.<sup>5</sup> For anatomical reference, Figure 1 depicts a normal temporal bone from a donor with no history of ear diseases, and Figure 2 presents a case of severe otosclerosis resulting in complete stapes fixation, which did not undergo surgical treatment.

Initially, there was little understanding of the actual progression of otosclerosis.<sup>4</sup> The establishment of otopathology laboratories, dedicated to studying the microscopic aspects of the temporal bone, facilitated further advancements in the field. Otopathologic examinations of human temporal bones not only described the underlying pathology, mechanisms, locations, patterns of progression, and inner ear changes caused by otosclerosis, but also contributed to the development of surgical strategies aimed at addressing the consequences of the bone remodeling (Figure 2). Initially, operations like the stapes mobilization were proposed to alleviate the symptom of fixation caused by otosclerosis. However, it became evident that simple mobilization of the stapes only yielded short-term relief, as the stapes often became fixated again, typically within weeks or months.

As otopathology gained prominence, a deeper understanding of the pathophysiology emerged, leading to improved comprehension of the disease state and the development of newer surgical techniques.<sup>7</sup> Otopathology has also been instrumental in elucidating the causes of failure and complications in stapedectomy and stapedotomy



**FIGURE 1** A representative temporal bone section at the middle ear level from a donor who did not have a history of ear diseases or histological evidence of middle or inner ear pathologic changes. (1) Incus, (2) middle ear, (3) facial nerve, (4) stapes footplate, (5) saccule/utricle, (6) cochlea, and (7) internal auditory canal (IAC).



**FIGURE 2** A representative human temporal bone specimen from a 91-year-old female who suffered from bilateral otosclerosis and who began developing bilateral hearing loss by the age of 12. Audiograms taken in her 50's indicate profound bilateral hearing loss, though no surgical intervention was performed. The fenestral otosclerosis progressed across the footplate to cause a complete oval window obliteration. There is a second otosclerosis foci located medial to the cochlea, with associated erosion of the anterior wall of the internal auditory canal.

procedures, such as poor prosthesis connection, round window obliteration, and hydrops, as it allowed access to the inner ear, which is not possible in vivo. Even today, human temporal bone pathology remains essential for understanding otosclerosis. Insights gleaned from otopathology studies have enabled the identification of mechanisms for sensorineural hearing loss resulting from otosclerosis, comprehension of complications associated with cochlear implantation in otosclerosis patients, correlation of clinical otosclerosis observed on CT scans with histopathology slides, and comparison of different

surgical techniques.<sup>8</sup> Additionally, the limits in postoperative imaging reduces the ability to assess correct prosthesis placement and surgical complications, so otopathological analysis can be crucial to understanding these complications. By translating findings from these studies into clinical practice, clinicians can further refine techniques and optimize patient care.

In this historical review, we aim to detail the progression of surgical techniques developed for otosclerosis through the lens of otopathology, providing histopathological images of exemplary and complicated cases for each technique. While the histology of the disease state has been well characterized, the histopathology of the array of associated interventions has not. By examining procedures through the lens of otopathology, we can gain unique insights into the history and evolution of stapes surgery.

## 2 | METHODS

From the Paparella Otopathology and Pathogenesis Laboratory at the University of Minnesota, we selected 8 specimens (ages 51–91) from donors with otosclerosis who had undergone surgical procedures using four methods (mobilization, fenestration, stapedectomy, and stapedotomy.) We also included one human temporal bone from the collection at the University of California Los Angeles that featured a case of stapedotomy. The study was approved by the respective Institutional Review Boards (UMN: 0206M26181; UCLA: 10-001449 and #22-001587). Representative cases showing the procedures and potential complications were selected and described. For comparative purposes, one extreme case of otosclerosis without surgical intervention, and one normal case without otosclerosis were also selected.

The temporal bones included in this study had been removed at autopsy, fixed with formalin, decalcified with ethylenediaminetetraacetic acid, dehydrated, embedded in celloidin, and serially sectioned at a 20 µm thickness. Every 10th section was stained with hematoxylin and eosin and was analyzed using light microscopy histopathological methods to identify otosclerosis foci, lesions, and other abnormalities.

## 3 | RESULTS

### 3.1 | Mobilization

The first attempt to surgically treat stapes fixation and otosclerosis was stapes mobilization, pioneered by Kessel in 1878, which showed promising results when he mobilized the entire footplate.<sup>5</sup> In 1891, Miot reported outcomes from over 200 stapes mobilization procedures, with the majority achieving surgical success, and 74 out of 124 experiencing improved hearing.<sup>9</sup> However, in the early 1900s, Politzer, Siebmann, and several others condemned the procedure, leading to its abandonment for decades due to the risk of bacterial infection and short term efficacy. Many patients experienced re-fixation of the stapes within weeks to months, rendering it a temporary solution.

Rosen, in 1953, revived the stapes mobilization procedure by accident during a fenestration operation, as the patient reported immediate improvement of the hearing after he inadvertently bumped onto the stapes.<sup>2,5</sup> Despite Rosen's experience, surgeons were still cautious about the mobilization procedures because most reports described only short-term results, lasting only for days to weeks. It was only in the 1950s that surgeons began to investigate a more direct approach to treat the ankylosed stapes such as the stapedectomy, rather than bypassing it via fenestration.<sup>10</sup> It is worth noting that stapes mobilization, though rarely used to treat otosclerosis, is still used as a method to examine stapes mobility, and to free the stapes in cases of stapedial fixation by tympanosclerosis.

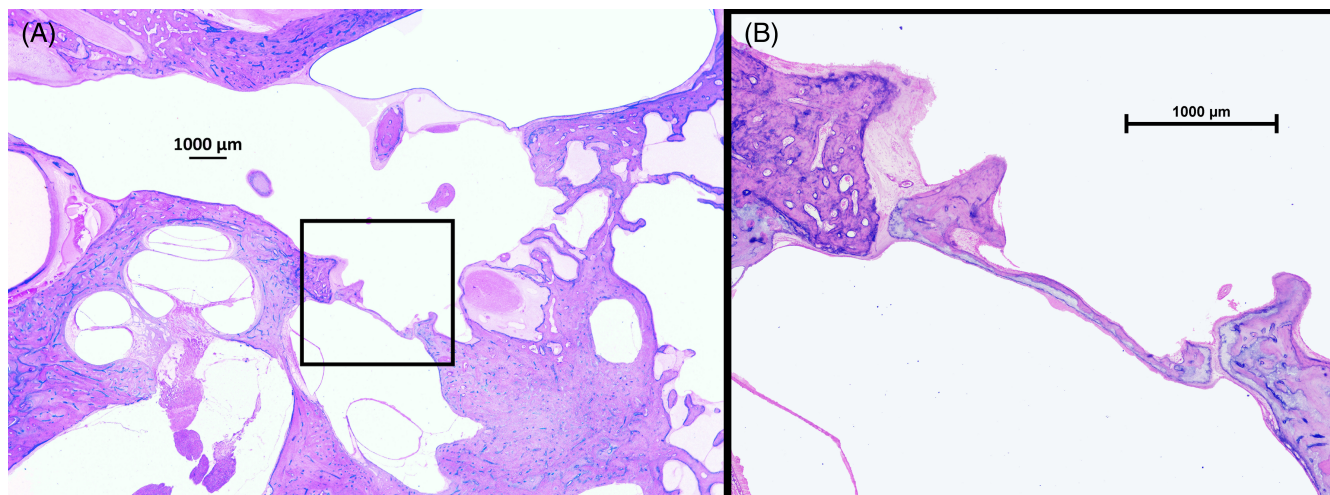
Another interesting note is that patients with a otosclerotic foci limited to the anterior aspects of the footplate along or a stapedial fracture outside of the foci had longer lasting hearing improvements.<sup>11</sup> As a result, several improved mobilization procedures have been attempted involving resection of the anterior crus and isolation of the otosclerotic foci. Recently, some physicians have returned to the concept of mobilization. Utilizing new laser and high-resolution endoscopy technology, Silverstein described the use of a laser stapedotomy minus prosthesis (STAMP) procedure to treat stapes fixation by mobilizing and isolating the posterior crus and posterior segment of the footplate.<sup>12</sup> Despite these findings, many surgeons still prefer the stapedotomy and stapedectomy today due to its success and low rate of re-fixation. Below, we present two temporal bones from a donor who underwent stapes mobilization surgery on both ears.

Figures 3 and 4 are representative of the results associated with stapes mobilization. These temporal bones are from a 90-year-old female who had bilateral stapes mobilization procedures to treat stapes fixation due to otosclerosis. The right ear (Figure 3) underwent a successful stapes mobilization procedure, whereas the left ear (Figure 4) experienced a complication resulting in the fracture of the footplate, which became displaced into the vestibule. Medical records indicate that the patient experienced hearing loss, but did not present with any other severe ear pathologies or symptoms. Audiograms revealed progressively worsening hearing loss. Unfortunately, the bone conduction thresholds were not performed or described in their audiogram, therefore we are unable to identify the type of hearing loss. Following mobilization surgery, the hearing thresholds in both ears temporarily improved, but subsequent audiograms showed progressive deterioration.

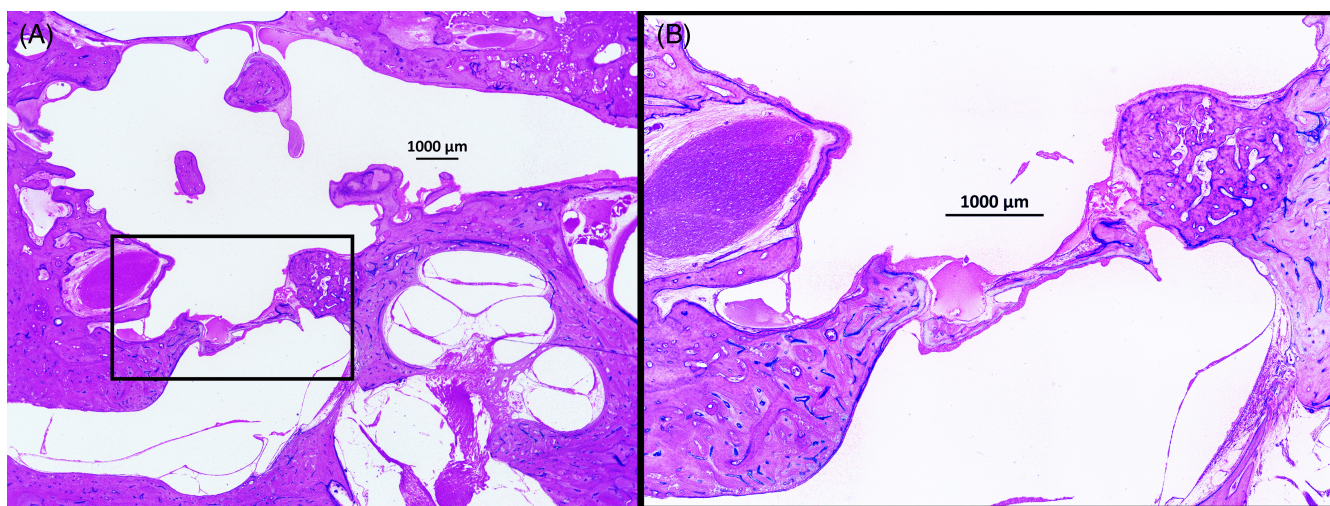
### 3.2 | Fenestration

The risks associated with stapes mobilization and its relatively short-term improvements led to the fenestration era. The first exploration of inner ear fenestration to bypass the fixed stapes footplate was described by Passov in 1897 and Floderus in 1899. They theorized that an opening in the promontory or vestibular labyrinth could bypass the fixed stapes, facilitating sound transduction.<sup>5</sup> In 1911, Barany observed improved hearing in an otosclerosis patient following an accidental fenestration of the horizontal semicircular canal during a





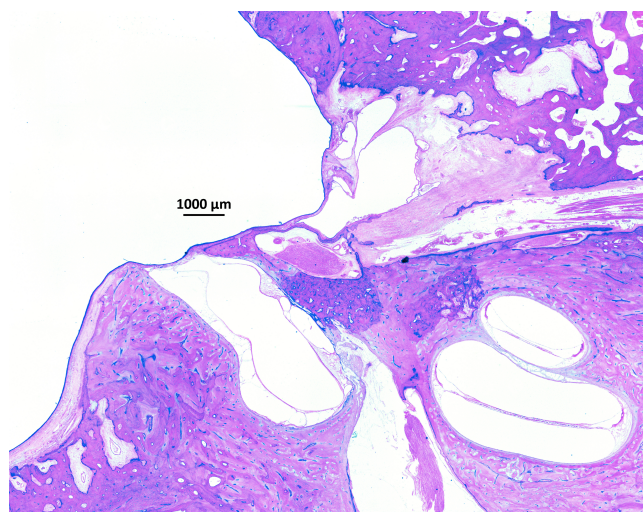
**FIGURE 3** (A and B) Two representative sections of a temporal bone from a donor who underwent stapes mobilization surgery (adjacent sections; A is directly superior to B). This is a good example of the short-term results associated with stapes mobilization. Although these specimens do not show any signs of stapes fracture or other major complications, it is possible to observe that the footplate is fixed in the round window. These findings are consistent with a lack of long-term benefit after the procedure.



**FIGURE 4** (A and B) A temporal bone section from the left ear of the same donor depicted in Figure 3. The pictures are representative of a complication of a stapes mobilization surgery. The histological analysis shows that the footplate was fractured and displaced into the vestibule. Even though the fracture occurred, the borders of the stapes seemed to have become separated from the otosclerotic focus, resulting in the temporary hearing improvement experienced by the patient post-operatively. However, the depicted section presents with fibrosis both on the anterior border of the footplate and between the pieces of the mid-shaft fracture, leading to re-fixation and the subsequent hearing deterioration. Furthermore, the displaced footplate into the vestibular organ may be the reason why the patient began suffering from unsteadiness and dizziness following the procedure.

radical mastoidectomy. However, it was not until Holmgren (1917) performed a series of otosclerosis fenestration operations, demonstrating that the inner ear could be safely accessed with sterile technique, that fenestration was considered a viable treatment option.<sup>2,13</sup> However, Holmgren's three-stage fenestration procedure took place over several months, and had limited results, leading to infrequent use of the surgery until Julius Lempert developed the single-stage fenestration technique in 1938.<sup>14</sup> In the procedure, the bone between the lateral canal and the membranous labyrinth is carefully removed, with

just a thin membrane layer remaining between the two structures, bypassing the stapes and oval window and creating a new communication into the inner ear. Lempert's innovation, coupled with his training of others in the procedure, resulted in widespread adoption and thousands of surgeries performed. However, outcomes were suboptimal for many patients, with a significant risk of postoperative complications such as sensory-neural hearing loss, extreme vertigo and nausea, ultimately leading to the decline of fenestration procedures.<sup>1,3,4</sup> Although the fenestration is now rarely performed,



**FIGURE 5** A representative human temporal bone from a donor who underwent a successful fenestration surgery. The picture shows a drilled mastoid with adequate re-epithelization. The lateral semicircular canal endosteum was opened, and the overlying epithelial membrane is intact without any signs of perilymphatic fistula. Otosclerosis is present near both the facial nerve and the cochlea and had completely encapsulated the stapes footplate (not shown), necessitating a fenestration procedure at the time. Post procedure records indicate that the procedure was successful in creating a 3rd mobile window without serious complications, and hearing was restored for the patient in this ear.

Lempert's surgical techniques paved the way for the development of future otologic practices, many of which are still in use today.

Figures 5 and 6 represent two different patients who underwent lateral semicircular canal fenestration surgery with different outcomes. Figure 5 is a temporal bone from a 80-year-old male who had bilateral otosclerosis who underwent a fenestration procedure at the age of 30 on the left ear and later a stapedectomy on the right ear. His audiograms reveal improved bone conduction thresholds following the procedure, which is representative of surgical success. On the other hand, Figure 6 shows signs of complications following fenestration. The temporal bone depicted in Figure 6 was from a 91-year-old female who had bilateral otosclerosis and complete stapes fixation in both ears, resulting in severe mixed hearing loss. She underwent bilateral fenestrations at the age of 30; post-operative audiograms do not show any improvements in the hearing thresholds for the right side due to complications.

### 3.3 | Stapedectomy

Dr. John Shea, in 1956, developed the stapedectomy procedure, in which he removed the stapes, covered the oval window with subcutaneous tissue, and inserted the then recently developed Teflon tube prosthesis and vein graft.<sup>15</sup> The procedure was a complete success and within the decade, became the standard operation for otosclerosis treatment. Though this procedure had been described in 1892 by Frederick L. Jack, and by Johannes Kessel (1876), this was the first

operation where the stapes was replaced with a prosthesis due to major advancements in magnification, surgical tools, biocompatible materials, and antibiotics.<sup>16</sup> After Dr. Shea's successful procedure, Schuknecht in 1960 created a steel wire-adipose tissue prosthesis, which helped to seal the vestibule, and connect to the rest of the ossicular chain.<sup>10</sup> Below, we present two temporal bones from patients who underwent stapedectomy procedures. The first case features a successful operation, with a correctly positioned prosthesis, and the second features an incorrectly positioned prosthesis, leading to complications.

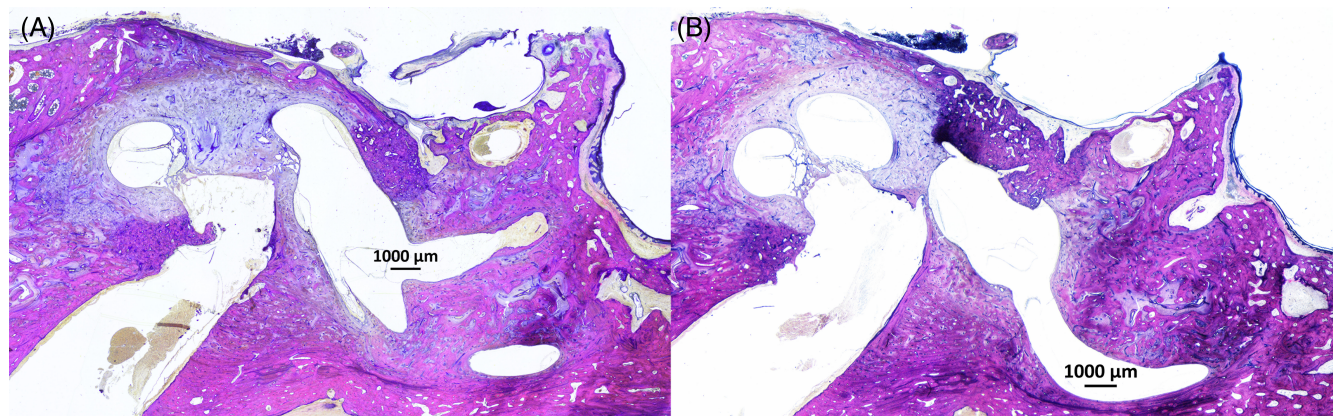
Figures 7 and 8 are from two different patients who underwent stapedectomy procedures, with the first case featuring a successful procedure, and the second featuring a potential complication from the operation. Figure 7 is a representative temporal bone section from an 87-year-old-female who had bilateral otosclerosis and presbycusis. She underwent bilateral stapedectomies to treat her stapes fixation. Comparison of left ear pre and post-surgical audiograms show a significant improvement of over 25 dB in all hearing thresholds after the stapedectomy operation. However, no bone conduction was measured. Conversely, Figure 8 features the temporal bone from a 51-year-old female with potentially serious stapedectomy complication where the prosthesis was inserted too far into the vestibule. She had progressive, bilateral hearing loss due to otosclerosis. She also sustained head trauma 1.5 years after her stapedectomy procedure, after which the patient suffered from sudden hearing loss. Pre and post injury audiogram comparison indicated a significant reduction in hearing across all frequencies.

### 3.4 | Stapedotomy

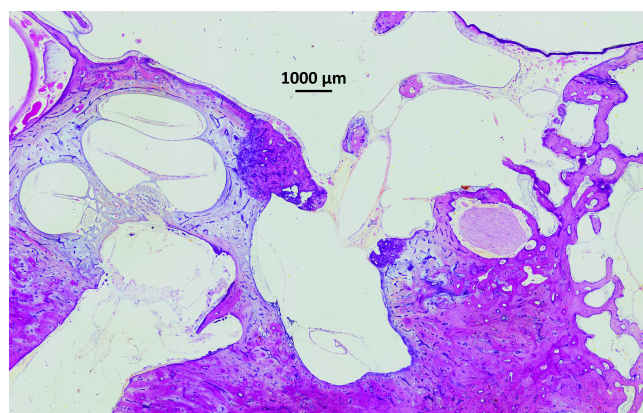
Eventually, as the surgery evolved, methods were developed to just remove a portion of the footplate sufficient to admit the prosthesis, known as a stapedotomy, leaving complete removal of the footplate for cases of greater severity. Stapes surgery is still evolving, with new tools and methods like the use of a laser to create the opening in the footplate currently being used.<sup>12</sup> This allows for a precise fenestration to fit the piston prosthesis and reduces the risk of injury to the vestibule and other structures. Below, we present two patients' temporal bones who underwent stapedotomy operations. The first case features a successful operation, with a successfully positioned prosthesis. The second case features two failed prosthesis and did not lead to improved hearing results due to the complications.

Figures 9 and 10 feature the temporal bones from two different patients who underwent stapedotomy procedures, with the first case being a successful operation, and the second displaying a potential complication. Figure 9 is from a 72-year-old male with bilateral otosclerosis which fixated the stapes footplate in both ears. No audiograms were present for this patient. The temporal bone featured in Figure 10 is from a female donor of unknown age. She suffered from severe hearing loss, and she underwent two tympanoplasties in an effort to close her large air-bone gap, with the second improving her thresholds by 10–15 dB. Later, she underwent two failed stapedotomies to further improve her hearing. After her first stapedotomy, she also reported severe vertigo due to rupture of the vestibular





**FIGURE 6** (A and B) Two representative human temporal bone sections from a donor with otosclerosis who underwent fenestration surgery. This patient experienced no improvement in hearing following the procedure. Histopathologic analysis shows an obliterative lesion in the oval window. There is evidence of drilling of the mastoid, with the membranous labyrinth of the lateral semicircular canal being open and communicating with the mastoid. Although there are no signs of fistula, the lateral semicircular canal has fibrosis and new bone formation, likely secondary to the fenestration procedure, which certainly further impaired sound transmission through the lateral semicircular canal. Although no information was found in the medical records, it is possible that the patient also experienced vestibular symptoms because of fibrosis.

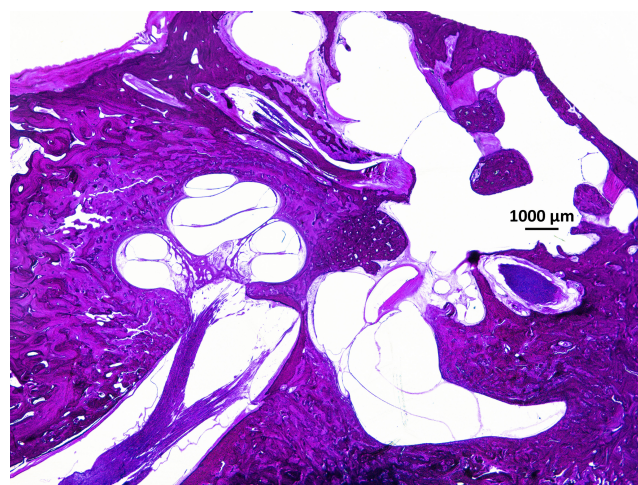


**FIGURE 7** Representative human temporal bones from a donor who underwent a successful stapedectomy operation. Otosclerosis had completely fixated both sides of the stapes, warranting a stapedectomy procedure, where the entire stapes is removed and replaced with a prosthesis, to yield good results. Much of the stapes footplate was removed, and a metal prosthesis was correctly implanted. While the implant was removed for temporal bone processing, the area where the implant would have been is shown by the void. The membranes that connect the prosthesis to the vestibule and inner ear are also intact. The audiogram indicated that the procedure was successful.

membranes and her audiograms showed profound hearing loss. Post second stapedotomy, the patient showed no improvement in hearing thresholds due to the incorrectly placed prosthesis as well.

#### 4 | DISCUSSION

The evolution of surgical techniques for treating otosclerosis has taken a long and complex path. From the pioneering attempts in the pre-antibiotic era, to the stapes mobilization in the late 19th century



**FIGURE 8** A representative temporal bone from a patient who suffered complications of the stapedectomy procedure. Otosclerosis had completely fixated the footplate, and a stapedectomy operation was performed. During the operation, the prosthesis (shown by the void) was inserted too far into the vestibule, perforating the membranous labyrinth and causing a labyrinthine fistula, leading to vertigo and perilymph leakage.

and the development of modern procedures like the stapedectomy and stapedotomy still used today, the field has seen significant advancements in safety and efficacy.

Stapes mobilization, once considered the best option, fell out of favor due to its short-term results, paving the way for fenestration as an alternative approach. However, it became evident that fenestration also had its drawbacks, including serious complications and lackluster results. The breakthrough came with the introduction of stapedectomy by Dr. John Shea in 1956, marking a significant advancement in otosclerosis surgery. This procedure, with subsequent refinements towards the stapedotomy, has become the current standard of care,



offering improved long-term outcomes and reduced risks for patients compared to the previous procedures.

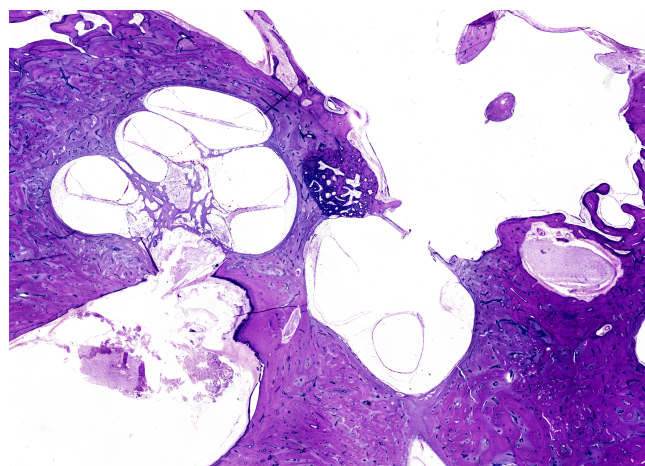
The future of otosclerosis surgical intervention holds several exciting possibilities. Advances in imaging technologies, such as high-resolution CT and MRI scans can provide surgeons with detailed pre-operative tools and can help enable personalized surgical planning.<sup>17</sup> Additionally, the integration of robotic-assisted surgery shows

promise in enhancing accuracy and eliminating tremor and drift.<sup>18,19</sup> Other innovations such as laser-assisted procedures offer increased precision and reduced risk, and recent studies have shown that endoscopic stapes surgery could be less invasive compared to traditional operations like microscopic surgery and may could improve visualization during operations.<sup>20</sup>

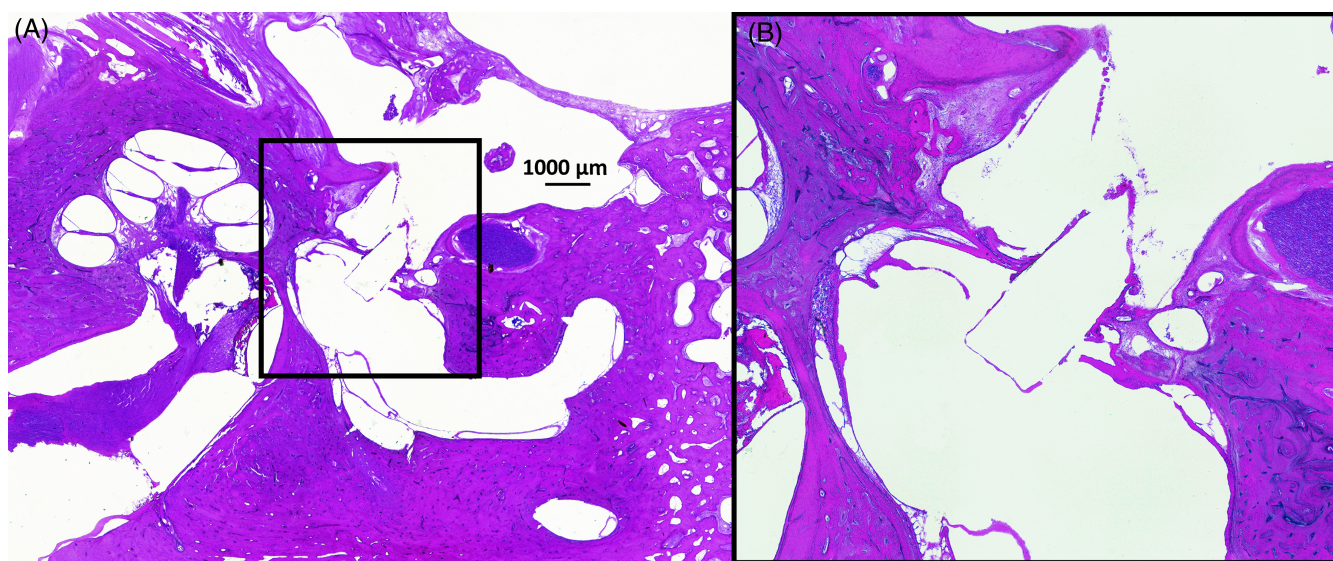
Studies published from animal models and otopathology research also play a vital role in advancing otosclerosis surgery techniques and analyzing their effectiveness. Animal models provide insights into the underlying pathophysiology of otosclerosis and can help evaluate the safety and efficacy of new surgical interventions. Otopathology studies offer valuable insights into the underlying pathological changes associated with otosclerosis and the effects of different surgical approaches on the pathology of the ear. There is also minimal postoperative visualization to identify correctly placed prosthesis and complications from surgery, so histopathological analysis of these cases postmortem can be critical to understand these complex cases. By translating findings from these studies into clinical practice, clinicians can further refine techniques and optimize patient care.

In the future, the addition of more specimens from donors with otosclerosis to otopathology collections may offer additional opportunities to explore recent techniques and innovations in the treatment of otosclerosis and stapes surgery. This future research could investigate the effects of laser fenestration and the histopathological correlates of bisphosphonate use in treating otospongiosis, for example.

In conclusion, while otosclerosis surgery has made improvements over the years, the field continues to evolve with the introduction of advanced techniques and technologies. With ongoing research and innovation, the future holds promise for further improving outcomes



**FIGURE 9** A representative temporal bone from a 72-year-old male patient who underwent a successful stapedotomy surgery. Otosclerosis had completely fixated the stapes, beginning at the anterior footplate, and progressing across the footplate. A central portion of the stapes was removed, and a Teflon prosthesis was correctly inserted into the removed portion, and no other complications or pathologies were observed.



**FIGURE 10** (A and B) Two representative sections of a case that demonstrates some of the potential complications resulting from a failed stapedotomy procedure. This case is unique because it features two incorrectly placed prostheses, both of which would not have improved the patient's hearing. As a note, both prostheses were removed during processing, but their outlines are present, indicating their position. The first prosthesis appears to have migrated out of the stapedotomy to rest on the residual posterior stapes footplate. The second prosthesis was positioned through the center of the footplate, but was inserted too deeply, displacing it into the vestibule. This placement likely caused her vertigo and fistula from ruptured vestibular membranes.

and quality of life for patients who require surgical intervention due to otosclerosis.

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## CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to disclose.

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