PHILIPPINE JOURNAL OF OTOLARYNGOLOGY-HEAD AND NECK SURGERY VOL. 38 NO. 2 JULY – DECEMBER 2023

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Diagnosis and Management of Otosclerosis in a Filipino Population: A Case Series

ABSTRACT

Objective: This case series aims to describe the diagnosis and management of otosclerosis in a series of Filipino patients encountered in a private, subspecialty otologic clinical practice. In particular, it elucidates the demographics, clinical presentation, diagnostic exam findings, clinical intervention, and hearing outcomes.

Methods:

Design: Retrospective review of medical records

Setting: Subspecialty otologic / neurotologic clinical practice

Participants: Medical records of all patients diagnosed to have otosclerosis based on radiologic evidence of fenestral or retrofenestral otosclerosis and/or confirmed during surgical exploration of the middle ear during the period 2004-2017.

Results: Nine patients with otosclerosis were identified. Seven patients presented with bilateral, moderate conductive hearing loss while two had bilateral mixed hearing loss. The patients had a mean age of 36.7 (range 18-48) with mean age at onset of symptoms of 30 years (range 16.5-45). Seven patients were from the component cities of the National Capital Region, one from the Cordillera region and one from south-central Mindanao. Radiologic evidence of fenestral otosclerosis was identified in 3 patients, of retrofenestral otosclerosis in 2 patients, while readings of normal temporal bone CT findings were seen in 4 patients. Obliterative otosclerosis was identified in one patient. Seven patients underwent stapedectomy (5 unilateral, 2 bilateral), and all seven had either objective or subjective evidence of hearing improvement post-operatively.

Conclusion: Clinical otosclerosis currently occurs in the Filipino population. It typically presents in middle age as a bilateral primarily conductive hearing loss. A younger age does not preclude its diagnosis, as juvenile otosclerosis has been identified. Identification of this condition in patients outside of the National Capital Region implies that it has to be considered even in regional populations without a racial predilection for otosclerosis. CT imaging of the temporal bone is essential in making the diagnosis, and should be part of the work-up in patients with conductive hearing loss and normal otologic examinations. Surgical treatment via stapes surgery is an effective and viable option in the management of otosclerosis in the Philippines. Specialized surgical equipment and appropriate surgical training is needed to deal with less common variants like obliterative otosclerosis.

Keywords: *otosclerosis; conductive hearing loss; Filipino; stapes surgery*

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The author declared that this represents original material, that the manuscript has been read and approved by the author, that the requirements for authorship have been met by the author, and that the author believes that the manuscript represents honest work.

Disclosures: The author signed a disclosure that there are no financial or other (including personal) relationships, intellectual passion, political or religious beliefs, and institutional affiliations that might lead to a conflict of interest.

Philipp J Otolaryngol Head Neck Surg 2023; 38 (2): 11-16

Otosclerosis is a focal osteodystrophy of the human otic capsule and stapes footplate, wherein abnormal bone remodeling (bone resorption, vascular proliferation, new bone deposition) occurs in the temporal bone. It typically presents in mid-adult life as a bilaterally progressive conductive hearing loss, although sensorineural hearing loss and mixed hearing loss may also occur. It is most commonly seen in Caucasians, and is considered rare among black, American Indian and Asian populations.[1,2,3]

Despite this relative rarity, a significant number of cases have been reported in Asian populations.[4-14] In contrast, there is a dearth of reported cases of otosclerosis in the Philippines. This is despite the fact that Filipinos share a common racial ancestry with the inhabitants of many of the countries[15] from which these reports have originated. Two imaging case reports were recently published by Yang.[16,17] Aside from this, a search of HERDIN Plus, the ASEAN Citation Index (ACI), Global Index Medicus - Western Pacific Region Index Medicus (WHO GIM-WPRIM), Directory of Open Access Journals (DOAJ), MEDLINE (PubMed and PubMed Central) and Google Scholar using the following search terms "otosclerosis", "Filipino", "Philippines" yielded only three reports on six cases[18,19,20] that were published more than half a century ago.

In addition, unlike in the United States, surgery for otosclerosis is not among the key indicator procedures for otolaryngology – head and neck residency training in the Philippines.[21,22] As such, it is not a condition which Filipino otolaryngologists are routinely trained to manage. Consequently, it would not be surprising if contemporary otolaryngologists in the Philippines fail to consider, or even totally discount the diagnosis of otosclerosis in patients with a clinical presentation compatible with this condition. In this light, there is a need to draw the attention of otolaryngologists to otosclerosis in Filipinos, by providing updated information regarding this condition in the Philippines, based on contemporary cases encountered in a subspecialty otologic practice.

This case series aims to describe the diagnosis and management of otosclerosis in a series of Filipino patients encountered in a private, subspecialty otologic clinical practice. In particular, it elucidates the demographics, clinical presentation, diagnostic exam findings, clinical intervention and hearing outcomes.

METHODS

With University of the Philippines Manila Research and Ethics Board approval (UPMREB Code 2023-0444-01), a retrospective chart review of all patients in a private subspecialty otologic practice with otosclerosis was performed to examine the presentation and treatment outcomes. Considered for inclusion were patients diagnosed to have otosclerosis based on radiologic evidence of fenestral or retrofenestral otosclerosis and/or confirmed during surgical exploration of the middle ear, during the period 2004-2017. Those with incomplete data (clinical or surgical records) would be excluded.

We based the radiologic diagnosis of otosclerosis on visual identification of any of the following findings on high-resolution computerized tomographic imaging of the temporal bone: a hypodense area of demineralization involving the enchondral layer of the otic capsule in the region of the fissula ante fenestram (fenestral otosclerosis), or the presence of a pericochlear hypodense area of demineralization that produces a "double-ring sign" of the cochlea (retrofenestral or cochlear otosclerosis).[23,24] All CT scans were obtained using helical multidetector scanners and were reviewed by the author.

We based the surgical confirmation of otosclerosis on palpation of the ossicular chain during transcanal middle ear exploration, with physical determination of immobility of the stapes and mobility of the malleus and incus, with or without visual identification of the otosclerotic focus in the area of the anterior oval window.[25]

The following data were obtained from the medical records: age, sex, city of residence, nature and duration of presenting symptom, prior treatment, audiologic test results, radiologic findings, surgical findings in those who underwent surgical management, and outcomes in relation to hearing status. We tabulated all data and analyzed it with relevant descriptive statistics.

RESULTS

We diagnosed nine patients with otosclerosis during the study period. At the time of diagnosis, their ages ranged from 18 to 48 years, with a mean age of 36.7. Four of the patients were male (44.4%) and five were female (55.5%). All of the patients presented with a history of bilateral hearing loss, with symptom duration ranging from 1.5 to 14 years. The age at which symptoms began to appear ranged from 16.5 to 45 years, with a mean age at onset of 30 years.

Seven patients were based in various component cities of the National Capital Region. One was a native of the Cordillera region (Baguio City), and one was a native of south-central Mindanao (South Cotabato).

Seven patients had bilateral moderate conductive hearing losses on audiometry, while two had bilateral moderate mixed hearing loss. Seven of the nine patients had tympanometry data, and all of them showed type A tympanograms.

Four patients were already managed with bilateral hearing aids, while one was advised hearing aid usage but declined. One patient was advised to seek treatment out of the country, while three did not have any prior treatment.

Temporal bone CT scans showed normal findings in four patients, fenestral otosclerosis *(Figure 1)* in three patients, and retrofenestral otosclerosis *(Figure 2)* in two patients. Those with "normal" temporal bone CT scans were diagnosed early in the series, when imaging studies were still printed on paper or photographic plates. In these studies, the images could not be magnified, nor their windowing parameters adjusted to ideal settings. As such, it is possible that these factors contributed to the inability to properly identify any subtle otosclerotic foci. The patients later in the series had DICOM imaging data on disks which could be evaluated on computers using magnified views and ideal windowing parameters for temporal bone imaging. In these studies, radiologic evidence of otosclerosis could be clearly identified.

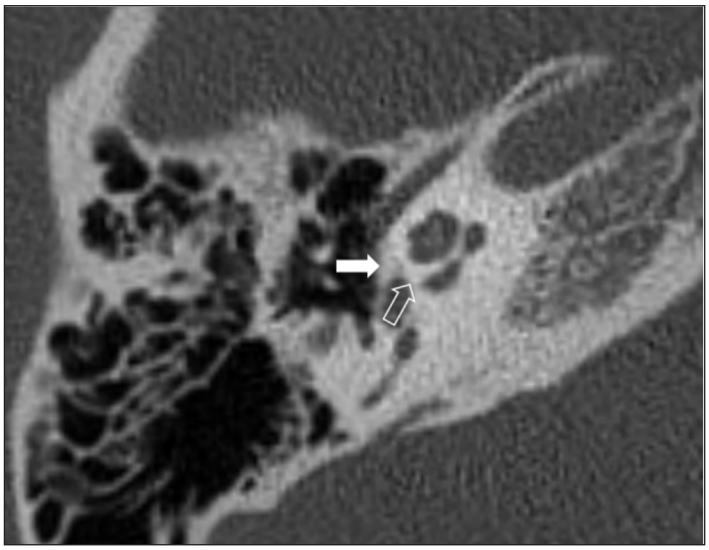

Figure 1. Temporal Bone CT, Axial slice, right side, Patient #8. Image at the level of the stapes footplate demonstrates the hypodense area of demineralization (white solid arrow) in the region of the fissula ante fenestram consistent with fenestral otosclerosis. This can be contrasted with the more solid bone (white outlined arrow) of the normal otic capsule.

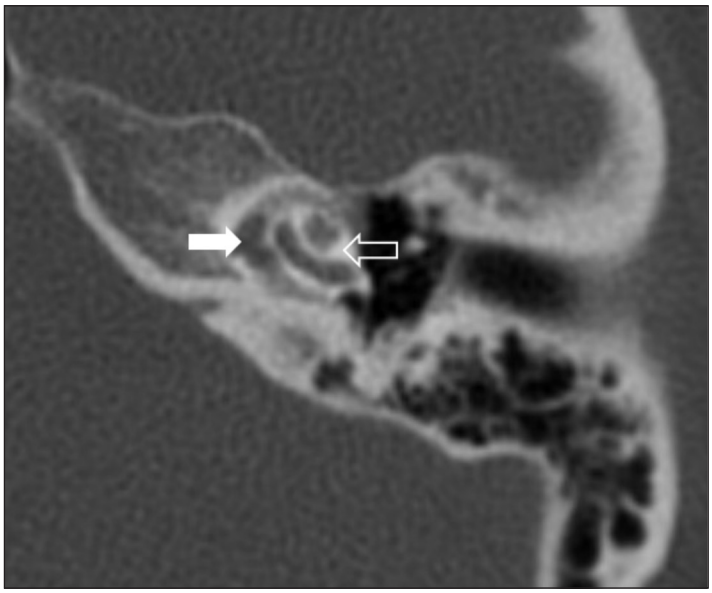

Figure 2. Temporal Bone CT, Axial slice, Left side, Patient #6. Image at the level of the basal turn of the cochlea (white outlined arrow) demonstrates the pericochlear hypodense area of demineralization (white solid arrow) that produces a "halo" or "double-ring" sign consistent with retrofenestral or cochlear otosclerosis.

Five patients had unilateral stapedectomy performed (three left-sided, two right-sided). Two patients had bilateral sequential stapedectomy. The technique of stapedectomy employed by the author for majority of the cases followed the general principles of stapedectomy with partial footplate removal and use of the Robinson bucket stapes prosthesis as previously described in the literature.[26] Teflon-wire piston prosthesis were used in Patient #1 and #3, as this was the only type of prosthesis available during that time. The KTP laser became available and was utilized for lysis of the stapedius tendon in patients later in the series. Patient #2, the youngest patient in the series, had bilateral sequential middle ear explorations, wherein obliterative otosclerosis was identified. *(Figure 3)* Stapedectomy was not performed due to the lack of appropriate instrumentation for safe footplate drilling at that time, and this patient was managed with hearing aids. In all other patients who underwent surgery, we identified otosclerotic foci in the region of the fissula ante fenestram located in the anterior portion of the oval window. *(Figure 4)* One patient declined to have stapes surgery, and continued to wear hearing aids in both ears.

Of the nine ears (7 patients) that had stapedectomy, two ears (one ear each in 2 patients) had near-complete closure (5 dB) of the air-bone (AB) gap. Four ears (both ears in 2 patients) had complete closure of the AB gap. *(Figure 5)* Three ears (one ear each in 3 patients) had markedly improved subjective hearing in the early post-operative period, with positive 512 Hz Rinne tests (AC>BC) on the operated side. Post-operative audiometry was not available for these three patients, as they had their subsequent follow-up with their referring physicians.

Of the four patients who were previously using hearing aids, 3 underwent stapedectomy (2 unilateral, 1 bilateral). All 3 ceased using hearing aids following surgery. One patient did not have surgery, and continued to wear hearing aids as his form of hearing rehabilitation.

With regards to surgical complications, one patient developed a floating footplate, and this fell into the vestibule. No attempts were made to remove the floating footplate once it had fallen into the vestibule. The patient had a marked improvement in hearing, but did have several episodes of positional vertigo during the first few post-operative months.

The demographic data, clinical presentation, audiologic and radiologic findings, and hearing outcomes are summarized in *Table 1*.

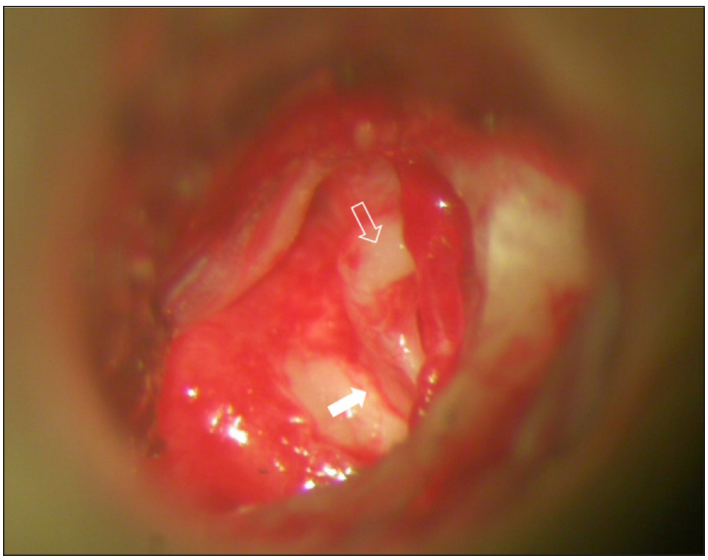

Figure 3. Intra-operative image, Patient #2. The otosclerotic focus has obscured the margins of the oval window (white solid arrow), such that the stapes crura appear to be arising from the otosclerotic focus. This is visual evidence of obliterative otosclerosis. The long process of the incus (white outlined arrow) serves as a landmark for the stapes.

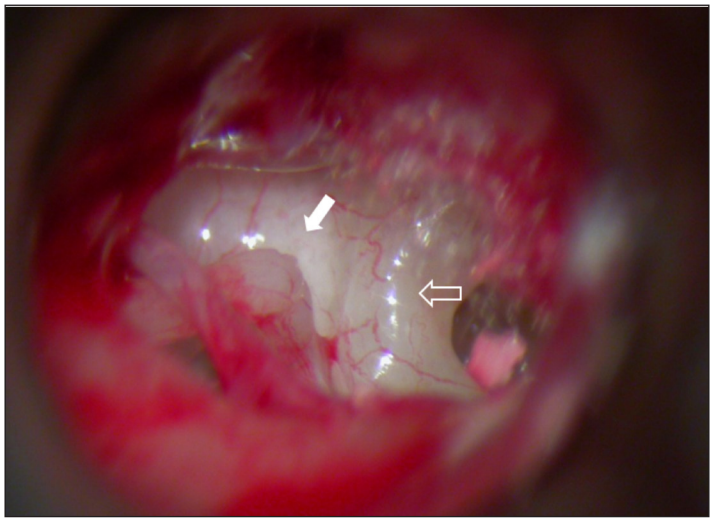

Figure 4. Intra-operative image, Patient #4. The bright white bone (white solid arrow) is the otosclerotic focus. This is reliably located in the anterior border of the oval window. This contrasts with the color of the surrounding normal bone (white outlined arrow)

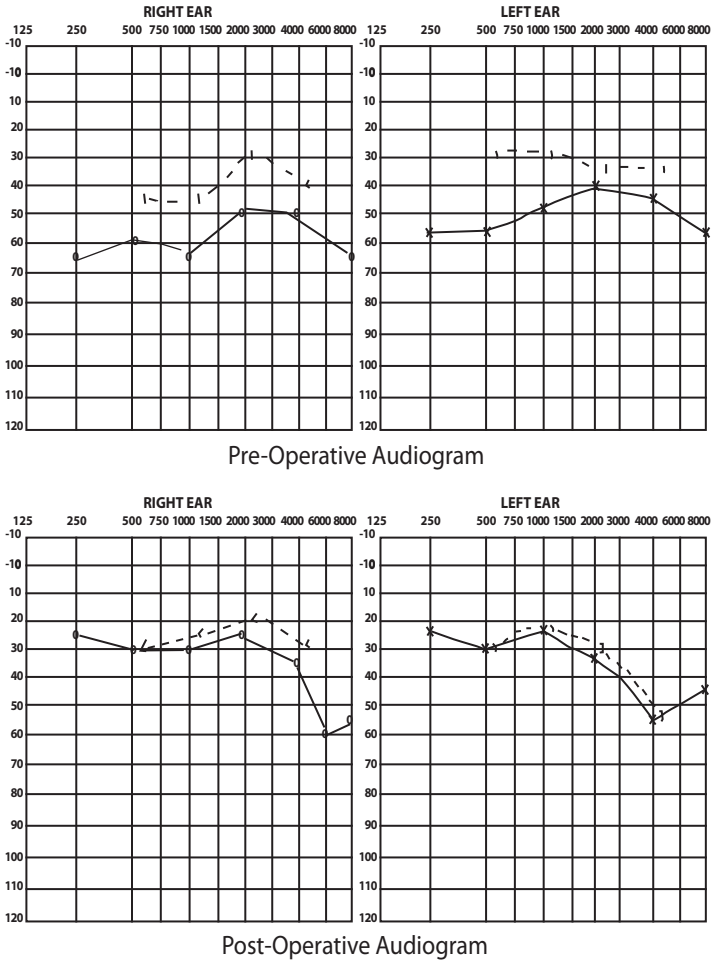

Figure 5. Pre-Operative and Post-Operative Pure Tone Audiograms (Patient #4) demonstrating total closure of the air-bone gap in both ears.

DISCUSSION

This case series is the largest to-date of Filipino patients with otosclerosis and provides important contemporary information about this condition in the Philippines.

First of all, most of the patients had clinical presentations typically seen in otosclerosis. Eight patients presented in middle-age with chief complaints of bilateral progressive hearing loss that were all primarily conductive in nature. All seven of the patients with tympanometric examinations had type A tympanograms. As such, encountering a middle-aged patient with bilateral conductive hearing loss and evidence of normal tympanic membrane and middle ear function should lead one to consider the diagnosis of otosclerosis.

However, younger age should not be a reason to discount the diagnosis. One patient (Patient #2) manifested with hearing loss at age 16, which places this patient in the distinct category of juvenile otosclerosis.[27] Patients in this category have a higher proportion of obliterative otosclerosis, where the footplate is greatly thickened, the oval window niche is filled-in to various degrees, and the annular ligament has been affected such that the margins of the oval window can no longer be delineated. In such cases, a burr is usually needed to penetrate the bone.[28] This was exactly the situation encountered in this young patient during surgery, where the procedures had to be aborted due to the unavailability of a microdrill that allows footplate surgery with much less risk of inner ear injury. This highlights the fact that despite its rarity, otosclerosis appears to present with a broad clinical spectrum

Table 1. Summary of Data

Patient	Age	Sex	City of Residence	Presenting Symptom	Audiologic Testing	Prior Treatment	Radiologic Evaluation	Management	Outcome
1	32	F	Baguio	BHL, 2 yrs	Moderate CHL	BHA	Normal TBCT	Left stapedectomy	Closure of AB gap to 5 dB left No longer wears hearing aids
2	18	M	Manila	BHL, 1.5 yrs	Moderate mixed HL	None	Normal TBCT	Right MEE Left MEE	Hearing unchanged. Wears hearing aids
3	26	M	Quezon City	BHL, 2 yrs	Moderate CHL Type A tympanogram	Advised Tx in USA	Normal TBCT	Left stapedectomy	Closure of AB gap to 5 db left. Expressed intention to operate right
4	47	M	Muntinlupa	BHL, 13 yrs	Moderate CHL Type A tympanogram	Advised HA	Normal TBCT	Right stapedectomy Left stapedectomy	Total closure of AB gap, bilateral
5	45	M	Surallah, South Cotabato	BHL	Moderate mixed HL Type A tympanogram	BHA	Retrofenestral otosclerosis on TBCT	Hearing aid	Hearing unchanged. Wears hearing aids
6	34	F	Quezon City	BHL	Moderate CHL Type A tympanogram	BHA	Retrofenestral otosclerosis on TBCT	Right stapedectomy Left stapedectomy	Total closure of AB gap, bilateral
7	45	F	Paranaque	BHL, 5 yrs	Moderate CHL Type A tympanogram	None	Fenestral otosclerosis on TBCT	Right stapedectomy	Markedly improved subjective hearing AC>BC at 500 Hz on operated side
8	48	F	Makati	BHL, 3 yrs	Moderate CHL Type A tympanogram	None	Fenestral otosclerosis on TBCT	Right stapedectomy	Markedly improved subjective hearing AC>BC at 500 Hz on operated side
9	36	F	Manila	BHL, 14 yrs	Moderate CHL Type A tympanogram	BHA	Fenestral otosclerosis on TBCT	Left stapedectomy	Marked improvement subjective hearing

BHL = bilateral hearing loss, CHL = conductive hearing loss, HL = hearing loss, HA = hearing aid, BHA = bilateral hearing aids, Tx = treatment, TBCT = temporal bone CT scan, MEE = middle ear exploration, AB = air-bone

in the Filipino population, including juvenile-onset and obliterative otosclerosis. Since these less common forms were encountered in such a small series, then the possibility of encountering patients on the other side of the hearing spectrum, that of far-advanced otosclerosis, cannot be discounted. This has implications on the diagnostic considerations in patients with what appears to be profound bilateral sensorineural hearing loss with unmeasurable air or bone conduction, as this may actually represent cases of far-advanced otosclerosis.[29] It also has implications in terms of the specialized surgical equipment that an otologic surgeon should have at one's disposal to prepare for the varied levels of pathology that may be encountered during surgery. The availability of microdrills for footplate fenestration and laser systems for small-fenestra stapedotomy will be relevant considerations when choosing the healthcare institution in which to perform surgery.

Secondly, most of the patients were residents of the National Capital Region. Although this may be attributed to the geographic proximity of the patients to the subspecialty otologic clinic practice where they were diagnosed and treated, one may hypothesize that the greater variety of racial backgrounds in the population of the National Capital Region may have contributed to the existence of patients with otosclerosis.

However, having one patient native to the Cordillera region, and another patient native to south-central Mindanao clearly indicates that the condition is also present in areas where the population may have a more homogenous, "native" racial background. This implies that one cannot dismiss the possibility of otosclerosis in patients with the classic clinical presentation simply because they do not have a racial background with a higher predilection for the disease (i.e. Caucasian race), and that it should be a consideration regardless of a patient's geographical region of origin.

Thirdly, high-resolution computerized tomographic imaging of the temporal bone is integral to the evaluation of patients with unexplained conductive hearing loss and the identification of otosclerotic lesions. The availability of the DICOM imaging data which can be viewed on a computer and manipulated to show magnified images with windowing parameters appropriate for temporal bone imaging is also a key factor in the proper identification of these lesions. Unlike the first four patients in the series whose imaging studies were on plates with unmagnified images and unmodifiable window settings, the last five patients in the series had DICOM imaging data which allowed the unequivocal visualization of both fenestral and retrofenestral otosclerotic foci.

Although not formally documented in this study, all of the imaging studies were read as normal by the attending radiologists. The identification of the otosclerotic foci on the radiologic studies were all done by the author, who had regular consultation sessions with a neuroradiologist while undergoing formal fellowship training in otology & neurotology. This situation highlights the need to increase awareness of this condition and its imaging characteristics in the professional radiologic community, and the current need to seek confirmation of the diagnosis of otosclerosis from those with experience in its identification.

Finally, surgical treatment of otosclerosis is an effective and viable management option for patients in the Philippines. However, due to the rarity of the condition and the highly technical nature of stapes surgery, a centralized referral system to clinicians with the training, dedicated interest, and constant surgical practice in otologic surgery[12,30] is integral to achieving optimal care.

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