Cochlear Fistula in Chronic Otitis Media without Cholesteatoma

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Cochlear fistula in the chronic otitis media (COM) without cholesteatoma is an extremely rare with only a few cases reported in the literature to this date. We describe a case of cochlear fistula observed in a female with COM without cholesteatoma. This report presents the first clinical case of a transtympanic iatrogenic trauma by habitual cotton swabs probably causing cochlear fistula.

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KEY WORDS: Cochlear fistula · Otitis media · Cholesteatoma.

Introduction

Cochlear fistula in the chronic otitis media (COM) without cholesteatoma is an extremely rare with only a few cases reported in the literature to this date [1-3]. We describe a case of cochlear fistula observed in a female patient with COM without cholesteatoma. This report presents the first clinical case of a transtympanic iatrogenic trauma by habitual cotton swabs probably causing cochlear fistula. Approval for this report was obtained from Inje University Sanggye Paik Hospital Institutional Review Board (IRB No. SGPIAK 2016-11-011).

Case Report

A 69-year-old woman presented at our hospital with 20-year history of right-side hearing impairment, otorrhea, ear fullness and tinnitus without vertigo. She habitually used cotton swabs for cleaning her ear due to frequent episodes of otorrhea and she also reported that she put cotton swabs deeply for some relief of ear fullness. She had a medical history of hypertension and diabetes mellitus. On physical examination,

a perforation of the tympanic membrane with granulation tissue was observed. The mucosa of the tympanic cavity especially above the promontory was very thick (Fig. 1). A pure-tone audiogram (PTA) showed right mixed type hearing loss with 55 decibels (dB) of bone conduction (BC) threshold and 90 dB of air conduction (AC) threshold. In comparison, the PTA in the left side was 36 dB of BC threshold and 46 dB of AC threshold (Fig. 2). Bacterial examination of the otorrhea subsequently yielded methicillin-resistant *Staphylococcus epidermidis*. High-resolution computed tomography (CT) was compatible with COM without cholesteatoma.

The patient went through surgery for COM via a postauricular approach. A canal-wall-up tympanomastoidectomy was performed. During the operation, a hard thickened mucosa was observed at the cochlear promontory. In removing the thickened tissue around promontory, above which fluctuation of the mucosa was noticed. An unconfirmed diagnosis of cochlear fistula was made and then CT was reviewed thoroughly. It showed bony defect of the first turn of the cochlea (Fig. 3). During the operation, the intact endosteum of the cochlea was noticed. Temporalis fascia was placed over the dehiscence of the cochlea. We did not find any other cholesteatoma lesions in the middle ear cavity. She did not complain of dizziness and further hearing loss after surgery.

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Fig. 1. Otoendoscopic view, right ear. The perforation of the tympanic membrane with granulation tissue and very thickened remnant of tympanic membrane and mucosa of the tympanic cavity especially above the promontory (arrow) with serous discharge were observed.

Discussion

Cochlear fistula in the COM without cholesteatoma is an extremely rare with only a few cases reported in the literature to this date [1-3]. The promontory is not usual site of bone destruction. It is very dense and not the common site of keratin accumulation and pressure necrosis even in COM with cholesteatoma unlike lateral semicircular canal.

In the COM with cholesteatoma, various mechanisms of bone erosion have been suggested. Besides pressure ischemia [4], other factors such as pH alterations, the activity of osteoclasts, production of osteolytic enzymes by the matrix of cholesteatoma, or a combination of the above been considered to play a role in bone resorption [5-7].

In this case of COM without cholesteatoma, repeated mechanical irritation such as habitual cotton swabs may cause chronic mucosal trauma and continuous inflammation in the cochlear promontory. Furthermore, inflammatory mediators induced bony remodeling in cochlear promontory. As a result,

Fig. 2. A PTA showed (A) mixed type hearing loss with 55 dB of BC threshold and 90 dB of AC threshold. In comparison, the PTA in the (B) was 36 dB of BC threshold and 46 dB of AC threshold. PTA: pure-tone audiogram, BC: bone conduction, AC: air conduction, dB: decibels.

Fig. 3. High-resolution axial (A) and coronal CT (B) scans of right temporal bone. CT scans shows severe mucosal swelling (arrows) above the destroyed bone covering the basal turn of cochlea (arrowheads).

those processes might lead to the cochlear fistula.

Sensorineural hearing loss in COM can be related to either mechanical or chemical irritations. Mechanical damage may include continuous trauma to the membranous labyrinth, leading to fistula formation between the endolymph and perilymph, causing a loss of hearing [8,9]. Chemical damage can be caused by the penetration of noxious substances such as bacterial toxins into the labyrinth. Endogenous toxins that may be present in middle ear effusions (lipopolysaccharides, prostaglandins, and leukotrienes) are also possible mediators of sensorineural hearing loss [10-13].

However, our patient's BC was relatively preserved. There are several hypotheses regarding the mechanism of hearing preservation. Natural segmentation of the labyrinth, a kind of anatomic adaptation, might prevent loss of endolymph [14].

When the cochlear endosteum was invaded by the cholesteatoma or was damaged during surgery, severe hearing loss occurred [14].

In the reported cases, the membranous labyrinth was left intact with preserved BC [15], even though membranous labyrinth conservation is not always synonymous with hearing preservation.

Chronic irritations by mechanical stimulation and inflammatory mediators may also cause local changes around the fistula such as thickening the endosteum of the cochlea and protecting involvement of membranous labyrinth. But further irritation at the endosteum could cause the hearing loss. A thick mucosal layer covered the unexpected small bone dehiscence of the promontory and, fortunately, its removal did not cause a lesion of the endosteum, allowing preservation of BC.

However, further evaluation is needed to find exact pathologic mechanism among mechanical irritation, cochlear fistula and hearing preservation.

In conclusion, cochlear fistula can also be observed in COM without cholesteatoma. It cannot be emphasized enough that CT should be read thoroughly preoperatively even in the COM without cholesteatoma case. The presence of a fistula is not always associated with severe hearing loss, so every effort should be made to not damage the membranous labyrinth. Inflammatory tissue or cholesteatoma matrix can be removed from the fistulous tract without injuring the membranous labyrinth. If any doubt of damaging the membranous labyrinth exists, the

matrix must be exteriorized and long-term follow-up is strongly recommended.

Therefore, even in COM without cholesteatoma, tympanoplasty would be recommended for preventing cochlear damage.

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Conflicts of interest

The authors have no financial conflicts of interest.

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