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CASE REPORT

Transplanted Cholesteatoma after Traumatic Tympanic Membrane Perforation

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Objective: To report an extremely rare case of transplanted cholesteatoma after traumatic tympanic membrane (TM) perforation.

Materials and Methods: The patient was a one year and four month-old boy who accidentally bruised his left temporal region against the hard ground. Left TM perforation with fresh bleeding and swelling of the auricle were identified.

Clinical observation with otomicroscopy and CT and then surgical treatment were performed. One month after the trauma, we confirmed that TM had spontaneously closed with thin membrane. Six months after the trauma, an isolated mass in the tympanic cavity was identified through closed area of TM. Intraoperative view showed that an isolated cholesteatoma existed without continuation with the TM.

Results: We considered that the pathogenesis of present case could be explained as following, the TM was initially blown open medially due to pressure increase in the external auditory canal and epithelial tissue was transplanted to the tympanic cavity. Then, the TM perforation spontaneously closed and an isolated cholesteatoma occurred without continuation with the tympanic membrane.

Conclusion: This case suggested that transplanted epithelium of TM has the potential to induce cholesteatoma in the tympanic cavity.

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Introduction

There have been some case reports of posttraumatic middle ear cholesteatoma, but these described limited cases of retraction cholesteatoma arising from temporal bone fracture or the secondary acquired cholesteatoma occurring from the edge of tympanic membrane (TM) perforation or intra-tympanic membrane cholesteatoma. We report a rare case of transplanted cholesteatoma after traumatic TM perforation.

Case Report

During the case management, the current ethics standards were taken into account. The patient was a one year and four month-old boy who accidentally bruised his left temporal region against the hard ground. His medical history was negative for otitis media, and there was no history of myringotomy or the other surgical treatment. After intracranial lesion was ruled out, he was referred to our department. Left TM perforation in the anterior-superior quadrant with fresh bleeding and

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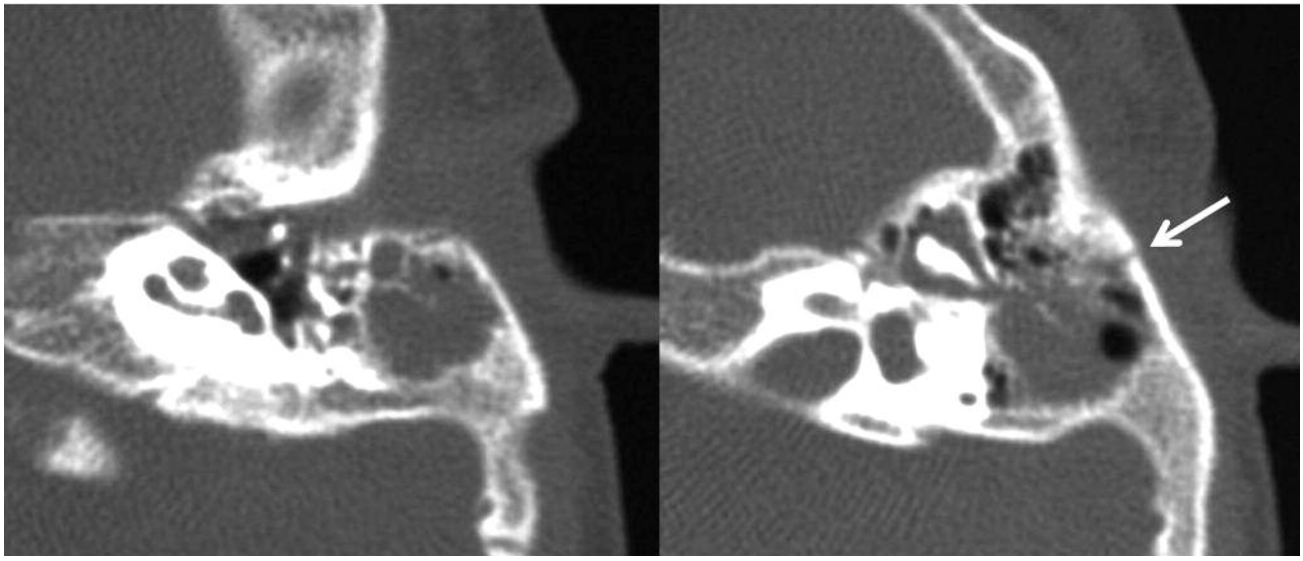

Figure 1. Axial temporal bone computed tomography performed immediately after the trauma demonstrated soft tissue density in the middle ear cavity and a minor fracture line on the surface of temporal bone (arrow). There was no other fracture or ossicular dislocation.

swelling of the auricle were identified. There was no facial nerve palsy and no nystagmus. There was no bone fracture involving the external auditory canal (EAC), middle ear ossicles, inner ear or facial canal, however soft tissue density in the middle ear and a minor fracture line on the surface of the temporal bone were identified on computed tomography (CT) (Fig. 1). One month after the trauma, we confirmed that TM had spontaneously closed with thin membrane. Six months after the trauma, an isolated white mass in the tympanic cavity was identified through the thin part of the TM (Fig. 2). CT scan demonstrated an isolated soft tissue density in the anterior part of the tympanic cavity without other abnormal shadow (Fig. 3). Conditioned orientation response audiometry with binaural examination showed normal hearing level. Eleven months after the trauma, extirpation of the lesion was performed under general anesthesia. After elevation of posteriorly-pedicled tympanomeatal flap with intrameatal incision, an isolated cholesteatoma was identified. The lesion was located on the mucosa of the tympanic cavity without continuation to the TM and was easily removed (Fig. 4). Pathological examination showed that most of the specimen was comprised of keratin debris with macrophages and some severely degenerated squamous epithelium was found. Neither subepithelial connective tissue nor infiltration of inflammatory cell was identified (Fig. 5). After the surgery, no recurrent lesion has been found for 3 years and normal hearing level in bilateral ears was confirmed with pure tone audiometry at the age of 5.

Discussion

There are many descriptions of posttraumatic cholesteatoma, but many of these reports described retraction cholesteatoma arising from the fracture line after temporal bone fracture[1,2]. They frequently occurred

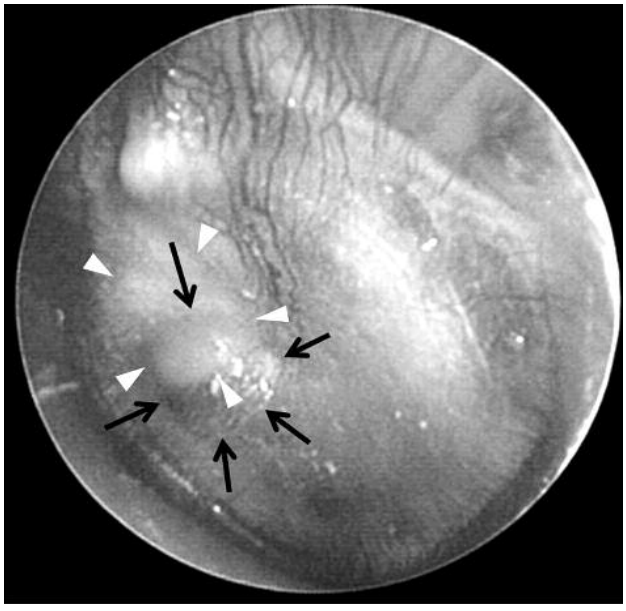

Figure 2. Otomicroscopy six months after the trauma showed spontaneously closed area on TM (black arrows) and an isolated white lesion in the tympanic cavity (white arrow heads).

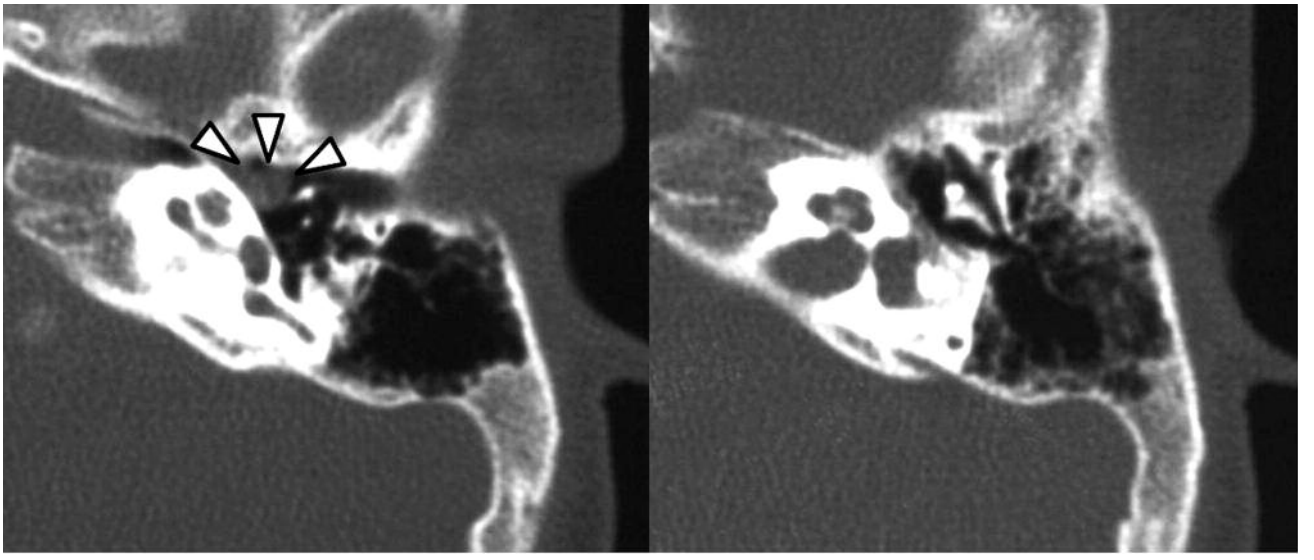

Figure 3. Axial temporal bone computed tomography six months after the trauma demonstrated an isolated soft tissue density in the tympanic cavity (arrow heads). The other soft tissue density in the middle ear cavity disappeared.

in the EAC, mastoid cavity and skull base. In the present case, there was no relationship between the epithelium of cholesteatoma and bone fracture, because there was no fracture line identified in the EAC and tympanic bone. However, a slight fracture line existed on the surface of temporal bone. Reports of cholesteatoma occurring after TM perforation were limited to cases of the secondary acquired cholesteatoma invading from TM perforation[3] or cases of intra-tympanic cholesteatoma occurring during the healing process of TM perforation[4]. In cases of secondary acquired cholesteatoma, epithelial invasion from the edge of TM perforation to the medial side of TM has been identified. In cases of intra-tympanic cholesteatoma, an epithelial pearl exists in the TM. The pathogenesises of those cases differ from the present case because cholesteatoma in the present case was isolated in the tympanic cavity.

On the other hand, we commonly encounter cases of congenital cholesteatoma with isolated lesion in the tymapanic cavity. But it is reasonable to suppose that the presented case was not a congenital cholesteaoma because the lesion was not identified in the tympanic cavity when TM perforation occurred, but was found through thin membrane after TM was closed spontaneously.

We considered that the pathogenesis of present case could be explained as following. Initially, TM was blown open medially due to a pressure increase in the EAC and epithelial tissue was transplanted to the tympanic cavity. Secondary, the TM perforation spontaneously closed. Then an isolated cholesteatoma developed without the continuation with TM.

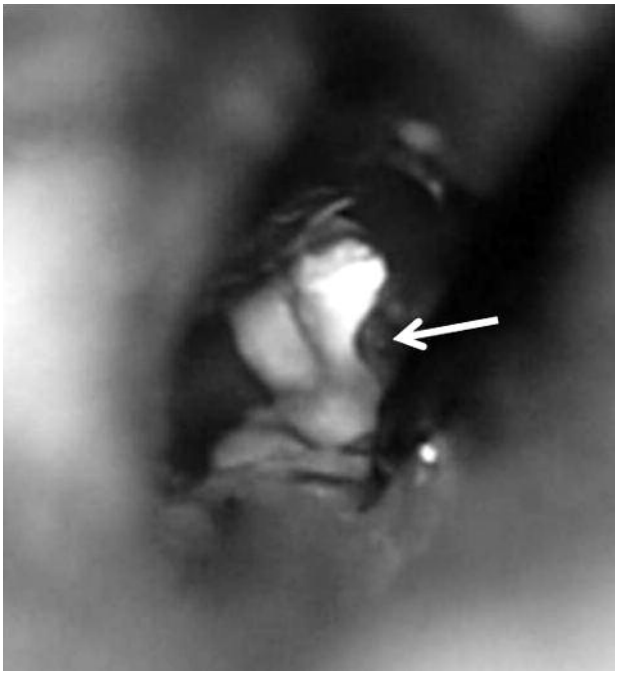

Figure 4. Intraoperative view showed that a cholesteatoma existed without continuation with the TM. Arrow indicates short process of the malleus.

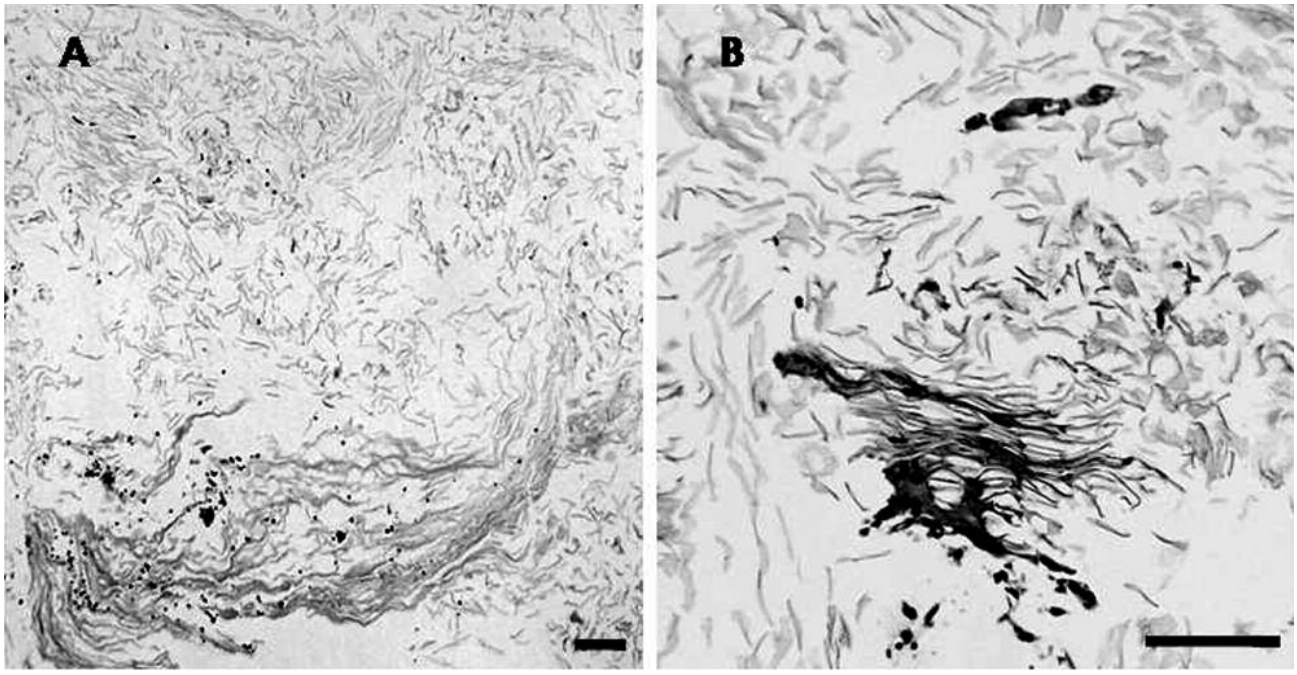

Figure 5. Pathological specimen (HE stains) was comprised of keratin debris (**A**) and severely degenerated squamous epithelium (**B**). Neither subepithelial connective tissue nor infiltration of inflammatory cell was identified . (scale bars indicate 100 μm)

In experimental studies using animal models, there were some reports describing transplanted cholesteatoma. Vennix et al described that transplanted meatal skin grafts were tolerated in the middle ear of the rat and superimposed infection arose with its expansive growth[5]. Hinohira et al also produced the transplanted cholesteatoma in the guinea pig and reported that exposed debris of transplanted epithelium plays an important role in the generation of inflammatory granulation[6].

In the present case, cholesteatoma formation was confirmed only six months after the injury. Initial inflammation caused by trauma and keratin debris might have induced rapid growth of the cholesteatoma. However, in the surgical specimen obtained eleven months after the trauma, there were no inflammatory cells but severe degeneration of the epithelium was identified. We speculated that the choleateatoma activity had decreased following the decrease in inflammation in the middle ear cavity after the TM perforation closure.

Conclusion

An extremely rare case of transplanted cholesteatoma after traumatic TM perforation was reported. To our knowledge, this is the first report describing such an occurrence. Despite its rarity, physicians should be aware that the transplanted epithelium of TM has the potential to induce cholesteatoma in the tympanic cavity.

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