External ear canal cholesteatoma after ventilation tube insertion and mastoidectomy

Holesteatom spoljnjeg slušnog kanala posle umetanja aeracione cevčice i mastoidektomije

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Abstract

Introduction. Etiopathogenetically, there are two types of cholesteatomas: congenital, and acquired. Numerous theories in the literature try to explain the nature of the disease, however, the question about cholesteatomas remain still unanswered. The aim of the study was to present a case of external ear canal cholesteatoma (EEC) developed following microsurgery (ventilation tube insertion and mastoidectomy), as well as to point ant possible mechanisms if its development. Case report. A 16-year-old boy presented a 4-month sense of fullness in the ear and otalgia on the left side. A year before, mastoidectomy and posterior atticotomy were performed with ventilation tube placement due to acute purulent mastoiditis. Diagnosis was based on otoscopy examination, audiology and computed tomography (CT) findings. CT showed an obliterator soft-tissue mass completely filled the external ear canal with associated erosion of subjacent the bone. There were squamous epithelial links between the canal cholesteatoma and lateral tympanic membrane surface. They originated from the margins of tympanic membrane incision made for a ventilation tube (VT) insertion. The position of VT was good as well as the aeration of the middle ear cavity. The tympanic membrane was intact and of normal appearance without middle ear extension or mastoid involvement of cholesteatoma. Cholesteatoma and ventilation tube were both removed. The patient recovered without complications and shortly audiology revealed hearing improving. Follow-up 2 years later, however, showed no signs of the disease. Conclusion. There could be more than one potential delicate mechanism of developing EEC in the ear with VT insertion and mastoidectomy. It is necessary to perform routine otologic surveillance in all patients with tubes. Affected ear CT scan is very helpful in showing the extent of cholesteatoma and bony defects, which could not be assessed by otoscopic examination alone.

Key words: cholesteatoma; tympanic membrane, perforation; ear, external; tomography, x-ray computed; reoperation.

Apstrakt

Introduction

First report of epithelial debris accumulation in the external ear canal was made in 1850 by Toynbee\(^1\). The term external ear canal cholesteatoma (EEC) was introduced in 1893 by Scholefield\(^2\). We presented a case of EEC after a ventilation tube (VT) insertion and mastoidectomy. It is a demonstration of a direct relationship between the use of a VT after mastoidectomy and the later development of canal cholesteatoma.

Case report

A 16-year-old boy presented with a 4-month history of a sense of fullness in the left ear and occasional otalgia on the same side. A year before admission the patient was submitted to surgery on the left ear due to acute purulent mastoiditis. Mastoidectomy and posterior atticotomy were performed with a ventilation tube (Tübingen-gold) placement. The patient was treated with antibiotics and his recovery was uneventful. On recall examination, microotoscopy was normal, as well as auditory and vestibular function.

On admission otoscopy examination revealed a complete obliteration of the left external ear canal (Figure 1). Pure tone audiogram showed left sided medium conductive hearing loss. There was no otorrhea or vertigo. Threedimensional multislice computed tomography (3D MSCT) showed a circumferential soft-tissue mass completely filling the external ear canal with associated erosion of the subjacent bone (Figure 2). A ventilation tube was clearly seen, with well aeration. A tympanic membrane was intact and of normal appearance without middle ear extension or mastoid involvement of cholesteatoma (Figure 3). The middle ear osseicules were also unaffected.
The patient underwent revision mastoidectomy surgery and extirpation of a VT. Intraoperatively, we confirmed normal findings in mastoid and middle ear cavities (Figure 4). Firstly, obliterations canal cholesteatoma was excised en block and then we removed a VT. From the margins of the tympanic membrane incision we noted a proliferation squamous epithelium remnants skipted laterally forming a connection with cholesteatoma. In the ear canal we also found the signs of bony wall erosion, particularly of the anterior wall. A defect of mental skin was excised and the irregular eroded area was drilled. The bone turned to normal and healthy canoplasty. Gross examination revealed an ovoid white shaped mass. Histopathology revealed stratified squamous keratinizing epithelial sac (matriks) with poor by developed perimatrix. Postoperatively, the patient was well. The place of tympanic membrane incision and mental skin were spontaneously healed within one week. Audiology showed improving in hearing with minimal loss around 20 dB. The patient was reviewed regularly and 2 years later showed no signs of the disease.

**Fig. 4 – The tympanic membrane is normal and anterior bony wall erosion (arrowheads) of the external ear canal is present after removing cholesteatoma (note the intact canal wall after mastoidectomy)**

**Discussion**

There are several well-known mechanisms of developing of EEC following different otologic surgeries. Cholesteatomas can occur in the anterior sulcus as a complication of lateral graft tympanoplasty. If the epithelium is trapped under the vascular strip, the cholesteatoma will form more laterally in the canal, along the posterior wall. Secondary EEC has been seen as a postoperative complication after mastoidectomy and may result from entrapment of squamous epithelial debris during the healing process.

Cholesteatoma is considered a complication of a VT placement when develops behind an intact drum or next to a perforation at or near the site of the tube insertion, in the mesotympanum or hypotimpanum. There is some controversy regarding the development of cholesteatoma following middle ear VT placement. Cholesteatoma may arise as a direct complication of a VT placement in a retraction pocket in an atrophic and flacid area in the drum that progresses to the point of debris accumulation, as a result of shedding and implantation of epithelial cells into the middle ear or due to ingrowth of squamous epithelium from the perforation margin to undersurface of the drum.

Theoretically, conditions for the development of secondary cholesteatoma are perfect both when VT is in place and when a perforation or atrophic scar remains at the implantation site. Because such cholesteatomas have been not encountered following myringotomy alone or mastoidectomy procedure we cannot accept seeding or implantation theories. Causative factor in our case could be reverse epithelial ingrowth from the incision margins to the undersurface of the drum, directed and enhanced by the tube’s flanges. Another possibility was mastoid infection stimulating changes in ear drum. Indication of mastoidectomy and of tube placement was usually chronic persistent middle ear effusion unresponsive to medical therapy. But in our patient VT insertion was performed due to acute suppurative mastoiditis. A bacterial infection initially could cause erosion of the epidermis layer and granulation tissue at the place of a VT and be partly related to later canal accumulation of keratin debris. But it is contradictory to EEC developed in an ear without recurrent episodes of otorrhea in a so-called “dry” ear without infection, as in case we presented. Most areas of focal atrophy or retraction are cosmetic and nonprogressive, related to the absence of a fibrous middle layer of the tympanic membrane at intubation site. This becomes problematic only rarely if progresses to a retraction pocket and onward to cholesteatoma (more common with longterm tubes). As spontaneous extrusion did not occur, and because ventilation was sufficient, intention to remove the tube was not realised. We assumed that cholesteatoma may not develop either in a retraction pocket, in an atrophic scar as the tympanic membrane was rather normal. We also hypothesised that as mastoidectomy have performed, the pattern of epithelial migration of external ear canal to outside could be influenced by restoring the normal ventilation of the middle ear. Slower migration rates have already been demonstrated in the inferior wall in patients with ECC and cholesteatoma we presented was mostly at that site. It was similarly suggested that it could be explained by hypoxic conditions due to poor blood supply.

The main symptom was progressive conductive hypoacusis and it may be related to occlusion of the external canal by cholesteatoma plug in oblitative cholesteatoma of ECC, but many cases can be remarkably silent or even asymptomatic. The invasion of squamous tissue with periostitis may explain chronic dull pain experienced by our patient. However, acute severe pain found more frequent in keratosis obturans. A preoperative high-resolution temporal bone CT is helpful whenever the surgeon suspects EACC eroding into adjacent anatomic structures. Erosion involving more than one ECC wall is typical but EEC could be more extensive than that suggested by clinical findings. For localized small...
lesions, treatment consists of frequent cleaning with debridement of necrotic tissue. Deeper pockets can be managed with canaloplasty by removing diseased skin and bone and exteriorizing the recess.

**Conclusion**

Although tympanostomy tubes are safe and efficacious for most patients with refractory otitis media or mastoiditis, they are associated with significant sequelae like cholesteatoma development. EEC is a rare entity after VT insertion with characteristic imaging and clinical features but different variables may influence its development. Exact fine mechanisms of cholesteatoma forming in the external ear canal near a tube placement site are still unknown. It is necessary to perform routine otologic surveillance in all patients with tubes. Affected ear CT scan is very helpful in showing the extent of cholesteatoma and bony defects, which could not be assessed by otoscopic examination alone.

**REFERENCES**


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