Otolaryngology–Head and Neck Surgery (2006) 134, 1055-1056

CASE REPORT

A new case of synchronous primary external ear canal cholesteatoma

Ricardo Persaud, MRCS, Arvind Singh, MRCS, Christos Georgalas, MRCS, Claudia Kirsch, MD, and **Michael Wareing, FRCS (ORL-HNS),** London, United Kingdom

In external ear canal cholesteatoma, there is excessive accumulation of keratin material in the external auditory canal as well as osteonecrosis with the focal appearance of sequestrated bone lacking an epithelial covering. The condition is rare, with an estimated incidence of 1 in 1000 new otological patients.[1] Here we present a unique case of bilateral primary external ear canal cholesteatomas in an otherwise healthy young African male and discuss the etiopathogenesis and management of the disease.

A 34-year-old African man presented with a 3-month history of a persistent dull ache in the left ear and a sensation of fullness in both ears. There was no tinnitus or otorrhea. Otoscopic examination revealed plugs of wax and squamous material in both ears. After microsuctioning of the left ear, a localized defect in the inferior part of the external auditory canal, anterior to the tympanic membrane, was noted. Squamous debris and loose fragments of bone were removed from the defect, revealing a large area of exposed tympanic bone. The remaining bony meatus was normal. Surprisingly, examination of the right ear revealed a similar but shallower defect. Both tympanic membranes were intact and normal in appearance. Pure tone audiograms and tympanograms were normal. A CT scan of the left temporal bone revealed a small soft-tissue irregularity extending inferiorly into the mastoid with sclerosis and bony destruction (Fig 1A). Similar findings were present on the right side (Fig 1B). No abnormality was seen in the middle or inner ear on either side. The radiological findings were consistent with the diagnosis of bilateral synchronous external ear canal cholesteatoma.

The initial management was conservative with regular aural toilet. However, the persistent dull pain in the patient's left ear became intolerable, and he opted for surgery. A postauricular approach was used to elevate the inferoposterior meatal skin exposing the disease in the ear canal. Squamous debris and a cholesteatoma sac were removed carefully, and the irregular eroded area was saucerized until normal healthy bone was found. The margin of the ulcer in the meatal skin was excised, and a temporalis fascia graft was used to line the exposed bone before repositioning the meatal skin. The ear canal was packed with ribbon gauze impregnated with Bismuth Iodine Phosphate Paste. It took approximately 10 weeks for complete re-epithelization over the denuded area. The patient is being reviewed every 4 months.

DISCUSSION

This is the first reported case of synchronous bilateral primary external ear canal cholesteatomas in a young black male. External ear canal cholesteatomas may also occur as a consequence of an underlying obstructive condition (eg, congenital stenosis) or surgical procedures (eg, tympanoplasty). There are many reported series describing secondary cases but only a few case reports of primary unilateral cholesteatomas.[2,3]

Two main theories exist on the etiopathogenesis of primary external canal cholesteatoma. The first theory states that the condition may be the result of minor trauma to the skin deep in the canal by, for example, a fingernail, cotton swab, or hard cerumen.[4] This may induce localized periostitis, and subsequent proliferation and invasion of the affected bone by squamous epithelium from the ulcer margin may lead to the formation of a cholesteatoma. Bony erosion may be caused by proteolytic enzymes or inflammation

St. Bartholomew's Hospital, London, England.
Reprint requests: Ricardo A. P. Persaud, MPhil, MRCS, DOHNS, MIBIOL, 50 Waller Road, Telegraph Hill , London, SE14 5LA, United Kingdom.

doi:10.1016/j.otohns.2005.03.031

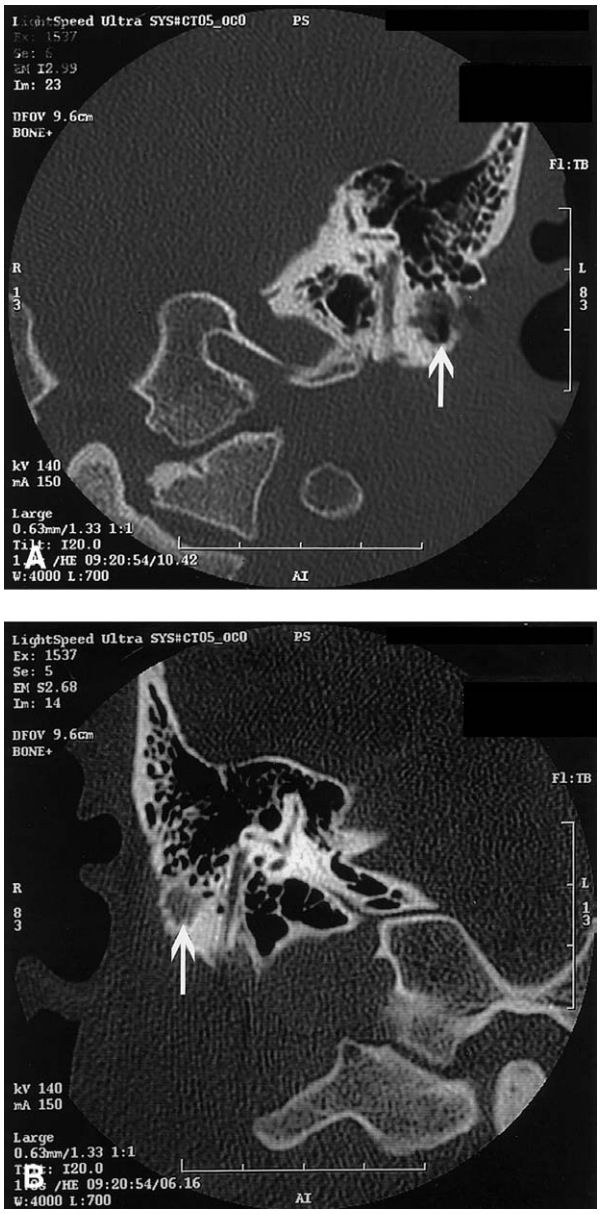

Figure 1 (**A**) Coronal CT scan of the left temporal bone showing a soft tissue irregularity with erosion in the adjacent bone in the floor of the external auditory meatus. **(B)** Coronal CT scan of the right temporal bone showing a soft tissue irregularity with erosion in the adjacent bone in the floor of the external auditory meatus.

associated with the cholesteatoma in a setup similar to that of the middle ear entity. The invasion of squamous tissue into a localized area of periostitis may explain the dull chronic pain experienced by affected patients (as in this case).

In the second theory, Holt[5] suggested that aging skin may lose its ability for proper epithelial migration and that aging cerumen glands may produce a drier wax that becomes more adherent to the ear canal skin, trapping desquamated epithelial cells. If the theory on migratory dysfunction is correct, then our case illustrates that the process may also occur in young patients.

The management of external ear canal cholesteatoma is debatable in terms of surgery versus conservative treatment. It has been recommended that all cases of the condition should undergo surgical removal of the cholesteatoma and necrotic bone to prevent progression and continued erosion.[1] The surgical procedure being determined by the extent of the osteonecrosis, erosion and the surgeon's own judgement. Investigators who have described secondary external ear canal cholesteatoma appear to agree with this recommendation, especially when (1) bony destruction extends into the middle ear or mastoid; (2) there is significant hearing loss or chronic discharge unresponsive to medical treatment; or (3) complications; such as facial nerve palsy and labyrinthine fistula, are present or imminent. Some investigators have suggested that treatment of the disease can be conservative as well as surgical, depending on the extent of the disease process.[2] The conservative approach may be suitable for small limited lesions and for patients with a contraindication to surgery or who refuse surgery.

In our patient, a conservative approach was adopted until the patient decided that the chronic left ear pain was intolerable and requested surgery. The surgical procedure was successful in eliminating the chronic ache from the left ear. It is likely that he will need a similar canal plasty on the opposite ear sometime in the future.

REFERENCES

1. Anthony PF, Anthony WP Surgical treatment of external auditory canal cholesteatoma. Laryngoscope 1982;92:70–5.
2. Vrabec JT, Chaljub G. External canal cholesteatoma. Am J Otolaryngol 2000;21:608–14.
3. Piepergerdes JC, Kramer BC, Behnke EE. Keratosis obturans and external auditory canal cholesteatoma. Laryngoscope 1980;90:383–91.
4. Bharadwaj VK, Walling KE, Rees J, et al. Necrosis and sequestration in the tympanic part of the temporal bone. J Otolaryngol 1984;13:299-04.
5. Holt JJ. Ear canal cholesteatoma. Laryngoscope 1992;102:608–13.