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Clinical Records

Atypical keratosis obturans

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Abstract
Keratosis obturans is a rare condition characterized by the accumulation of desquamated keratin material in the bony portion of the external auditory canal. Classically, it is reported to present with severe otalgia, conductive deafness and global widening of the canal. A case of keratosis obturans is described in which the principal symptom was a metallic taste and the main finding was extensive erosion of the hypotympanum with exposure of the facial nerve and the annulus of the tympanic membrane. This presenting symptom and resorption pattern are atypical of keratosis obturans and have not been documented previously.

Key words: Ear, External; Keratosis; Dysgeusia

Introduction

Keratosis obturans is a rare condition in which cholesteatoma-like material is found deep in the external ear canal. The term was first used in 1874 by Wreden when he presented 12 cases of 'a peculiar form of obstruction of the external auditory meatus, consisting of a compact mass of tissue, which is distinguished from an ordinary mass of cerumen.'[1] However, it was Toynbee who first described the condition in a case report published in 1850.[2]

The characteristic clinical manifestations of keratosis obturans are severe otalgia and conductive deafness secondary to accumulation of desquamated material in the ear.[3] There may also be circumferential widening of the bony canal.[4] In this case report we highlight an atypical presenting symptom and a pattern of bony resorption not described previously.

Case report

A 42-year-old woman was referred by her General Practitioner because of a strange metallic taste in her mouth and a vague feeling of fullness in the left ear. She did not complain of any other ear symptoms such as hearing loss, discharge, pruritis, pain or tinnitus. There was no significant past medical history. Examination of the oral cavity was unremarkable but otoscopic examination of the left ear revealed a large amount of keratin debris in the canal and only a small portion of the tympanic membrane was visible. Her audiogram was normal. After microsuction of the keratin material a large defect was noted in the hypotympanum and the facial nerve and annulus were clearly visible; the tympanic membrane appeared normal, although slightly hyperaemic (Figure 1). The cavity was lined with normal-looking squamous epithelium. There was no evidence of osteonecrosis or sequestrum formation. The right ear was normal. A diagnosis of keratosis obturans was made and the patient was seen again in six months. She has continued to accumulate sheets of keratin debris within the 'autohypotympanectomy cavity' and has required microsuction every six months.

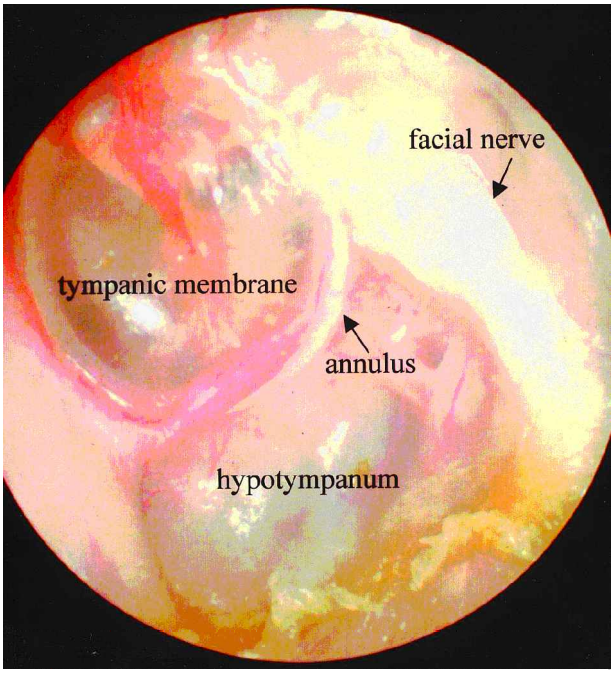

Fig. 1
Extensive erosion in the hypotympanum caused by keratosis obturans.

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Discussion

Until the late 20th century, keratosis obturans and external ear canal cholesteatoma were thought to represent the same disease and therefore the terms were used interchangeably to describe the accumulation of desquamated keratin in the bony part of the external auditory canal.[5] In 1980, Piepergerdes *et al*. reviewed the literature and concluded that keratosis obturans and external ear canal cholesteatoma are two different diseases requiring separate management.[3] However, while both conditions can occur independently, they do have some similar characteristics which can sometimes make it very difficult to make a precise diagnosis.[6] Classically, patients with keratosis obturans present with acute conductive deafness, and severe pain secondary to the accumulation of keratin in the ear, while external ear canal cholesteatoma is characterized by otorrhoea and a chronic dull unilateral ache secondary to the invasion of squamous tissue into a localized area of periostitis in the canal.[3] Keratosis obturans tends to be associated with sinusitis or bronchiectasis in 77 per cent of juvenile and 20 per cent of adult cases.[7] Bilateral occurrences are also more common in children.[8]

- **This paper is a case report describing keratosis obturans with recurrent dysgeusia each time the keratin re-accumulates**
- **The pathophysiology of keratosis obturans is discussed**

Here we present a case of unilateral keratosis obturans in an otherwise healthy adult in whom the main symptom was a strange metallic taste. Such a symptom may be associated with chorda tympani dysfunction after middle-ear surgery when the nerve may be stretched or damaged, but it has never been reported as a presenting symptom of keratosis obturans. It is our conjecture that the pressure exerted by the keratin plug on the exposed facial nerve may be responsible for the symptom as the patient reported an improvement following the removal of the keratin mass.

Naiberg *et al*.[9] stated that erosion of the underlying bone does not occur in keratosis obturans. However, Hawke and Shanker[10] reported a case of automastoidectomy caused by neglected keratosis obturans. In that case the bone resorption occurred just posterior to the bony annulus and extended back into the mastoid but no soft tissue structures were reported to be exposed. In our patient there was extensive bony erosion predominantly in the hypotympanum with exposure of the vertical part of the facial nerve and the annulus of the tympanic membrane; the jugular bulb was almost dehiscent (Figure 1). This pattern of resorption of the underlying bone, may be due to the pressure exerted by the expanding mass of keratin deep in the ear canal.[10] The disease appeared to spare the soft tissue structures.

The aetiology and the pathogenesis of keratosis obturans are not clearly understood. The cause has been related to eczema, seborrhoeic dermatitis or furunculosis,[11,12] as well as bronchiectasis, which causes reflex sympathetic stimulation of the cerumen glands and the subsequent formation of an epidermal plug.[7] Paparella and Shumrick[13] suggested that the epidermal plug may also be due to excessive production of epithelial cells or faulty migration. Normally, ink-dot markers placed in the centre of the ear drum moved laterally[14] whilst markers placed on the drum directly over the malleus handle moved upwards to the pars flaccida and then posterosuperiorly.[15] In keratosis obturans there may either be abnormal migratory pathways, no migration or an abnormally slow movement of epithelium leading to accumulation of squamous debris.[15–17] These abnormal processes may either affect the tympanic membrane alone or the external auditory canal giving rise to two forms of the condition; keratosis obturans of the tympanic membrane is thought to be the less severe form.[16] The pathogenesis of the abnormal migration may be associated with damage to the migratory basal epithelial cells following inflammation involving these cells and that a similar aetiology may be responsible for the inflammation of the lower respiratory tract and paranasal sinus that often co-exists.[17]

Hawke and Shanker[10] also suggested the existence of two different forms of keratosis obturans. In one form there is a chronic inflammation within the subepithelial tissues and this is responsible for the hyperplasia of the epithelium and the accumulation of keratinous material in the external canal. In the other form there is no inflammation in the skin lining the external canal. This latter form occurs bilaterally and may be due to a hereditary or acquired deficiency in an enzyme (not yet identified) which is responsible for the separation of the superficial layers of keratin. This must occur if these layers are to glide outwards during normal migration.[10] Whilst in our case the epithelial tissue appeared normal, there was no evidence of keratosis obturans in the opposite ear.

It has been suggested that keratosis obturans associated with chronic inflammation of the epithelium in the auditory canal may be cured once the keratin plug is removed and the underlying inflammation successfully treated.[10] However, patients with keratosis obturans not associated with any inflammation of the canal skin will require life-long cleaning of the ear at regular intervals because of the local metabolic deficiency affecting the normal migratory mechanism.[10] We opted to treat our patient with regular microsuction as she continued to accumulate keratin debris within the deep portion of the auditory canal. We found that this treatment was necessary every five to six months when the metallic taste returned. It is likely that our patient will require this treatment for the rest of her life.

Summary

We report a case of keratosis obturans that is atypical in terms of the main presenting symptom of a metallic taste and extensive bony erosion with dehiscent facial nerve and annulus of the tympanic membrane and we summarize the literature on the pathophysiology of the condition.

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