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CLINICAL RECORD

A rare case of primary otoscleroma of the middle ear

A R KAKERI, A H PATEL

Department of ENT, Al-Ameen Medical College, Bijapur, Karnataka, India

Abstract

Objective: We report an extremely rare case of primary otoscleroma.

Method: We present a case report and a review of the world literature concerning otoscleroma.

Results: An adult woman presented with chronic suppurative otitis media with tubotympanic disease and conductive hearing loss. On mastoid exploration, dark granulations were seen, which were identified as otoscleroma on histopathological examination. The patient responded well to streptomycin.

Conclusion: To the best of our knowledge, this is the first report of primary otoscleroma in the world literature. This case indicates that Frisch's bacillus can also spread to the middle ear.

Key words: Ear, Middle; Otitis Media; Rhinoscleroma

Introduction

Rhinoscleroma or scleroma is a progressive, granulomatous disease which commences in the nose and which can eventually extend into the nasopharynx, oropharynx and larynx.[1,2] It can sometimes affect the middle ear, hence the term otoscleroma. It is commonly seen in patients with poor hygiene[1] and in those of reproductive age.[2–4] Frisch's bacillus has a predilection for the upper respiratory tract.[2] It can also present at other associated sites, and with chronic suppurative otitis media with or without complications.[3]

Here, we present a rare and interesting case of otoscleroma.

Case history

A 34-year-old woman presented to the out-patient department of Al-Ameen Medical College Hospital, Bijapur, with a 7-year history of left ear discharge, with reduced hearing over the previous 2 years.

Examination revealed a large central tympanic membrane perforation with polypoidal changes in the middle-ear mucosa.

An X-ray of the mastoid (Schüller's view) revealed sclerosis.

Her hemogram was normal.

Pure tone audiometry revealed a 40 dB, conductive hearing loss.

The patient underwent mastoid exploration under local anaesthesia. Intra-operatively, abundant dark, polypoid masses and granulations were found within the mastoid antrum and middle ear, from which tissue was obtained and sent for histopathological examination. The malleus and the lenticular process of the incus were eroded and necrosed. Granulations were present over the facial canal. The lateral semicircular canal dome was identified and found to be intact. There was a greater degree of peri-operative bleeding compared with routine cases. After removing the diseased tissue, a tragal cartilage ossiculoplasty (inter position) was performed together with a temporalis facial graft.

Post-operatively, cefotaxime (1 g) was administered intravenously twice daily for 5 days. The post-operative period was uneventful.

Histopathological examination (Figure 1) revealed chronic granulomatous changes with the presence of Mikulicz cells, Russell bodies and macrophages, indicating rhinoscleroma.

The patient's nose, paranasal sinuses, oral cavity and larynx were examined but no abnormality was detected. A biopsy was taken from the nasal mucosa and turbinates but did not reveal any scleromatous changes. Streptomycin (0.75 g) was administered intramuscularly (after a test dose) once daily for four weeks.

Three months after surgery, the graft had taken well, there was a 15 dB hearing improvement, and the mastoid cavity was dry.

Discussion

Rhinoscleroma per se involves the nose; however, it sometimes only affects the middle ear, in which case it is termed otoscleroma. The latter location is very rare, with only two previous reports.[3,4] Sites such as the lacrimal sac,[6] larynx and trachea can also be affected.[5] Complications of otoscleroma have been reported: for example, Barbary *et al.*[3] reported otoscleroma with facial nerve palsy. However, in our case as well as that reported by Goravalingappa and Belagavi,[4] there were no complications. Rhinoscleroma spreads along the submucosal plane from the nose into the nasopharynx, the eustachian

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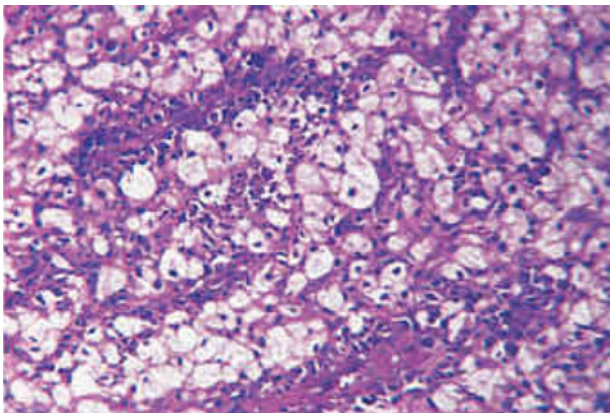

FIG. 1

Photomicrograph of otoscleroma showing macrophages, plasma Russell bodies and Mikulicz cells. (Hematoxylin–eosin stain ×200)

tube and the middle ear. The larynx, trachea and lacrimal sac[1,3–6] can also be involved.

- **Primary otoscleroma is very rare; usually the nose is involved (rhinoscleroma)**
- **This adult case of primary otoscleroma presented with otorrhoea and deafness**
- **Mastoid exploration showed dark granulation tissue**
- **Diagnosis was made histologically from mastoid granulation tissue**
- **The patient responded to streptomycin**

Occasionally, primary otoscleroma may be encountered without any evidence of disease elsewhere, i.e. the infective organism is inoculated directly into the middle ear,[4] either by droplet infection from the nose or nasopharynx, or through the external ear via tympanic membrane perforation.

Various post-operative hearing outcomes have been reported. In our case, hearing improved by approximately 15 dB, whereas previously reported patients experienced no improvement in hearing.[3,4]

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Address for correspondence:
Dr Ashfak R Kakeri,
Professor,
Department of ENT,
Al-Ameen Medical College Hospital,
Bijapur 586108,
Karnataka State, India

E-mail: ashfak_kakeri@yahoo.com

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