Cholesteatoma and tuberculosis: Association or coincidence

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ABSTRACT

Primary tuberculosis of the middle ear is often a diagnostic dilemma due to its rarity and variable clinical presentations. A chronic presentation is chronic otorrhoea with multiple perforations of the tympanic membrane. We report the case of a 41-year-old migrant worker who presented with a post auricular abscess without other otological symptoms. Radiological and histopathological findings revealed a cholesteatoma and tuberculosis.

Keywords: Cholesteatoma, tuberculosis, otitis media

INTRODUCTION

The tuberculosis bacilli (TB) was first isolated by Koch in 1882, but TB of the ear was only first described by Davidson *et al*. in 1960. [1] Primary TB of the head and neck most commonly manifests as cervical lymphadenitis. Tuberculosis rarely affects the ear and the disease is usually secondary to infection of the lung, pharynx, larynx and nose. [2] Tuberculous otitis media usually poses a diagnostic dilemma due to its rarity and variable clinical presentations. To date, tuberculosis in association with cholesteatoma has only been reported as a superimposed infection in one series by Yanif *et al*. [3]

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CASE REPORT

A 41-year-old gentleman, an migrant worker with no prior medical illness was referred to our centre with a right post auricular swelling for the past 15 years. This had suddenly increased in size, had ruptured and was discharging pus. Prior to this, he denied any symptoms of otorrhoea, otalgia or impaired hearing. There were no constitutional symptoms or history of tuberculous contact. Examination revealed a tender left post auricular swelling measuring about 3x3cm with a sinus opening over the superior part of the swelling. There was no facial asymmetry. Otoscopy showed sagging of the posterior wall of the external auditory canal which obliterated the view of the tympanic membrane. No obvious keratin debris was seen. There was no evidence of cervical lymphadenopathy. His pure tone audiometry showed right sided severe to

profound sensorineural hearing loss and normal hearing on the left side. Our clinical impression was right cholesteatoma with a mastoid abscess.

A plain and contrasted high resolution computed tomography (HRCT) scan showed a large soft tissue density with calcifications and destruction of right mastoid air cells. These imaging findings were suggestive of a right post auricular and mastoid abscess. An incision and drainage was performed at the referring hospital and the cultures isolated *Morganella morganii*. A Magnetic Resonance Imaging (MRI) scan of the temporal bone performed at the same centre showed features consistent with a cholesteatoma or chronic inflammation, which was in close proximity with the right temporal lobe, with no intracranial extension.

The patient's chest radiograph and Mantoux test were normal and his ESR was mildly elevated at 19mm/hr (<15 mm/hr). Screening for human immune deficiency virus (HIV) was negative. A repeat HRCT of the temporal bone with 1mm cuts was performed at our centre and this showed a large soft tissue density within the right mastoid and middle ear cavity, with extensive destruction of the ossicles, adjacent bone as well as sclerosis of the inner ear. With the clinical impression of a cholesteatoma, the patient underwent a right mastoid exploration. Examination under microscopy revealed a posterior sagging of the external auditory canal and the tympanic membrane appeared intact. A large cholesteatoma sac occupied the mastoid with erosion of the posterior wall and tegmen tympani. All the ossicles were absent. Based on these findings, a right radical mastoidectomy was performed.

Examination of the cholesteatoma specimen showed acid fast bacilli (AFB) on Ziehl Neelsen staining. There were no classical pathological characteristics of TB such as central caseous necrosis, epitheliod cells and Langhan's giant cells. The TB culture from the cholesteatoma was negative. The patient recovered well postoperatively and was referred to the Respiratory team for consideration of anti-TB therapy. He was commenced on Ethambutol, Isoniazid, Rifampicin and Pyrazinamide for six months. At the last follow-up about four months ago, he was asymptomatic and the right ear was dry.

DISCUSSION

TB is still one of the commonest infectious diseases which causes significant morbidity and mortality. Primary TB of the middle ear mainly occurs due to direct implantation through the external auditory canal and a perforated tympanic membrane. [4] Secondary TB usually occurs by aspiration of infected secretions of the upper respiratory tract with retrograde transmission or via haematogenous spread from other infective foci. [4] In the case described above, the infection was most likely secondary in view of an intact tympanic membrane and no history of otorrhoea and no other possible TB foci found.

Uncommon and non-specific presentations usually result in delayed diagnosis. The diagnosis is often only recognised postoperatively or when complications such as facial paralysis, labyrinthitis, post auricular fistulae, subperiosteal abscess and intra cranial extension of infection have occurred. [3] TB of the middle ear typically presents with painless

chronic otorrhoea with multiple perforations of the tympanic membrane that fails to respond to antimicrobial therapy. [4] Patients may also have conductive, sensorineural or mixed hearing loss. Most reports describe patients with tuberculous otitis media as having long-standing otorrhoea. [3] The case discussed above, however, did not display the classical features that have been described by other authors.

This patient had cholesteatoma with an intact tympanic membrane. Congenital cholesteatoma is unlikely as it usually presents at a younger age. There are few possible mechanisms of acquired cholesteatoma with an intact tympanic membrane. Firstly, a narrow-necked retraction pocket might have detached from the tympanic membrane which later healed leaving a keratin cyst behind it. [6] Another possibility is migration of squamous epithelium from the edge of a perforation followed by healing of the defect. [6]

Based on literature search, the only association made between tuberculosis and cholesteatoma was described by Yanif et al. [3] They reported six patients with cholesteatoma and tuberculous infection of the middle ear as sequelae to chronic suppurative otitis media. Diagnoses were established by histopathological examination. [3] All their patients also had positive cultures to other organisms. Microscopic examination for AFB is usually not a preferred test as it requires approximately 10^5 organisms to be present to obtain a positive result. [7] In our case, AFB was seen in the cholesteatoma and culture was negative.

Anti-TB chemotherapy is the mainstay of treatment for tuberculous otitis media. [8] Some authors suggest that surgical intervention is reserved for managing complications that may arise. [9] Other authors agree that surgical management is also useful in cases when there is poor response to anti-TB medications or to aid diagnosis. [8] Surgical intervention was warranted in this patient due to the clinical impression of cholesteatoma. The diagnosis of tuberculosis in this patient was made postoperatively. The question that arises is due to the fact that the bacilli were found confined within the cholesteatoma sac and the whole sac was removed during the surgery. As his TB screening were also negative, we were uncertain whether anti-TB treatment was warranted. Following consultation with a Respiratory physician, this patient was put on anti-TB regime for six months.

Based on this case, we recommend routine Ziehl Neelsen staining to detect acid fast bacilli of all cholesteatoma specimens from high risk workers. Further studies on TB within cholesteatoma may elucidate the pathogenesis, whether the bacilli are lying dormant within the sac due to low virulence or whether the cholesteatoma has encapsulated the bacilli and limited its spread.

In conclusion, TB of the middle ear is rare and the association with cholesteatoma is even rarer. Atypical presentations may pose a diagnostic dilemma. TB should be entertained as a potential differential diagnosis even with atypical presentations. Further studies of TB within cholesteatoma may elucidate its pathogenesis in the middle ear.

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