Unusual presentation of more common disease/injury

Tuberculous otitis media: a case presentation and review of the literature

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Summary

A 42-year-old farmer being treated for pulmonary tuberculosis was referred to our clinic after developing otorrhoea and hearing loss in his right ear. Examination revealed a large subtotal perforation in the right ear in addition to a purulent discharge with right facial nerve palsy. Audiometry indicated a 35 dB conductive hearing loss at 0.5, 1 and 2 kHz with air–bone gaps of 12, 15 and 10 dB, respectively. A middle ear biopsy was performed under direct visualisation, with a middle ear lavage which was positive for allergic fungal sinusitis (AFS). The final diagnosis was tuberculous otitis media (TOM).

Clinical symptoms and signs should be reviewed in every case of chronic otitis media keeping the possibility of tuberculosis in mind. Otorrhoea in a patient with known or suspected active pulmonary tuberculosis should be assumed to be TOM until proven otherwise.

BACKGROUND

Tuberculous otitis media (TOM) is a rare cause of chronic suppurative infection of the middle ear and mastoid. The incidence of tuberculosis in the middle ear is very low and accounts for only 0.04% of all cases of chronic suppurative otitis media. Its diagnosis is often delayed because it can easily be confused with other acute or chronic middle ear conditions.

CASE PRESENTATION

A 42-year-old farmer presented with a 6-month history of cough, productive of blood-stained sputum, weight loss and physical weakness. Four months later he developed otorrhoea and progressive hearing loss in the right ear. He initially presented to the pulmonology unit of our institution from where he was referred to our unit on the same day for co-management. Upon referral to our department,

Figure 1 A video-otoscopy image of an ear with a central perforation and purulent discharge.

examination revealed an inflamed and oedematous external auditory canal, a large subtotal perforation (figure 1) in the right ear and purulent discharge with right facial nerve palsy. Audiometry indicated a 35 dB conductive hearing loss at 0.5, 1 and 2 kHz with air–bone gaps of 12, 15 and 10 dB, respectively. Other systemic examination findings were normal.

INVESTIGATIONS

A middle ear biopsy was performed under direct visualisation, with a middle ear lavage which was AFS (allergic fungal sinusitis) positive using the Ziehl–Neelsen staining method. A final diagnosis of TOM was made. Three sputum smears from the patient were 2+ for acid fast bacilli (AFB). An aural swab for microscopy yielded *Pseudomonas* species, which was sensitive to ciprofloxacin. Chest x-rays revealed multiple irregular densities in both lung fields that were consistent with slowly resolving foci of tuberculosis. Serology for HIV was negative.

DIFFERENTIAL DIAGNOSIS

Differential diagnoses include Wegener granulomatosis, herpes zoster oticus, non-tuberculous otitis media, cholesteatoma, diabetes otitis externa, Lyme disease, HIV and infectious mononucleosis.

TREATMENT

Treatment was instituted with isoniazid, rifampicin, pyrazinamide and ethambutol for 6 months, and daily aural wick dressing with ciprofloxacin ear drops.

OUTCOME AND FOLLOW-UP

The ear discharge resolved after 3 months and the perforation of the tympanic membrane healed completely after 9 months, with an average 25 dB improvement in the hearing deficit at 0.5, 1 and 2 kHz. Complete resolution of the facial nerve palsy was observed after 1 year.

DISCUSSION

In developing countries including Nigeria, TOM is a very common infectious disease whose frequency has risen because of the AIDS epidemic.¹ It is caused by *Mycobacterium tuberculosis*, *Mycobacterium hominis* and *Mycobacterium bovis*, the latter two of which generally affect the ears (rare species of mycobacterial infections can sometimes cause atypical infection especially where there are immunodeficiencies). *M bovis* is seen less frequently than *M hominis*. TOM is usually due to ingestion of infected cow's milk.

Tuberculosis remains the leading cause of death from infectious disease worldwide in persons above the age of 5.² It is one of the major infectious diseases with predominant involvement of the lung and lymph nodes. However, tuberculosis of the middle ear is uncommon³ (accounting for only 0.04% of all cases of chronic suppurative otitis media in the UK^{4 5}) as in most cases the origin is a pulmonary focus and primary cases are rare.⁶

In the pre-antibiotic era, 2–8% of all cases of chronic suppurative otitis media were due to tuberculosis and 50% of cases were in infants less than 1 year of age.⁷ Very few cases of TOM are reported in the literature. Mills' study mentioned that the incidence of TOM has fallen dramatically

since the beginning of the last century.⁸ At that time, 3–5% of cases of otitis media were due to tubercle bacillus, whereas today the condition is rare,⁸ largely due to childhood immunisation programmes. Turner and Eraser reported in 1915 that 2.8% of all cases of suppurative otitis media were due to tuberculosis.⁷

The prevalence of active or inactive pulmonary tuberculosis in patients with TOM ranges from 14% to 93%.^{9–11} Conversely, about 2% of patients with active pulmonary tuberculosis have TOM.¹² TOM should be strongly considered in patients with known or suspected tuberculosis and a chronic ear infection; however, lack of evidence of tuberculosis elsewhere does not exclude the possibility of TOM.¹³

Histological findings are important for the identification of *Mycobacterium*, which shows granuloma formation with caseous necrosis, epithelioid cells and Langhans giant (multinucleated) cells.¹⁴ The middle ear may be filled with ectopic bone in which small tubercles, with their characteristic Langhans multinucleated giant, epithelioid and round cells, are found. The ossicles, except for the stapes footplate, may be absent. Tubercles often occupied the facial canal, in place of the nerve, just posterior to the geniculate ganglion.¹⁵ Three types of changes are frequently seen: miliary, granulomatous and caseous. The miliary type is associated with superficial infection, the granulomatous type with superficial bony involvement, and the caseous type with massive necrosis and sequestration.¹⁶

Tuberculosis can also be transmitted congenitally and is associated with a high incidence of ear involvement. However, congenital tuberculosis is extremely rare and hardly ever presents with isolated ear involvement.

The pathogenesis of TOM involves three major mechanisms. The first mechanism is aspiration of mucus through the Eustachian tube, the second is blood-borne dissemination from other tuberculous foci and the third is direct implantation through the external auditory canal and a tympanic membrane perforation.¹¹ TOM used to be more common in children than in adults possibly because the Eustachian tube anatomy of children permits reflux of material into the middle ear cavity.^{11 12}

The classic clinical features of TOM were described by Wallmer in 1953 as painless otorrhoea, multiple tympanic membrane perforations, pale granulation tissue, ipsilateral facial nerve paralysis, early severe hearing loss and bone necrosis.¹⁷ The multiple perforations in the early stages may later coalesce into a total tympanic membrane perforation accompanied by pale granulation tissue.^{5 11 18}

However, these classical features are rarely observed today. Recently a review of all reports of TOM in the English literature by Skolnik *et al* refuted these findings. Their research showed that facial palsy is present in only 16% of cases and multiple tympanic perforations are equally rare.¹²

In the early stages of TOM, the drum looks dull and some dilated vessels can be observed.¹⁹ The tympanic membrane then becomes thickened and landmarks are obliterated.¹⁷

The otorrhoea described as being painless, can actually be painful^{9 11} due to granulation tissue in the middle ear and possible bacterial superinfection. In the series described by Nishiike *et al*, none of the patients had multiple perforations, facial nerve palsy or bone erosion.²⁰

Generally tuberculosis of the middle ear is unilateral.^{14 21} This is surprising as one would expect it to be bilateral since a possible route for middle ear infection is through the Eustachian tube. Associated facial nerve paralysis is seen in approximately 16% of adult cases and 35% of paediatric cases.²²

Involvement of the temporal bones by tuberculosis was first described by Jean Louis Petit in the 18th century, and the clinical signs of the disease were first outlined by Wilde in 1853. Koch demonstrated tuberculous bacillus in 1882 and Esche isolated bacillus in middle ear secretions in 1883.^{23 24}

External ear canal cultures have been reported to be positive for tuberculosis in 5–35% of cases, and smears are positive in approximately 20%.²⁵ However, confirming the diagnosis can be difficult because the high rate of secondary bacterial infection of the tuberculous middle ear (79%) can prevent the identification of *M tuberculosis* on either staining or culture.^{25 26 27}

Early diagnosis and prompt treatment may prevent ear damage and central nervous system complications.

Pure tone audiogram can reveal conductive, sensorineural or mixed hearing loss. Conductive hearing loss is seen early in the disease when there is tympanic membrane perforation. With the onset of labyrinthitis, mixed hearing or sensorineural hearing loss occurs as corroborated by MacAdam and Rubio who reported a case of slowly developing hearing loss, suggesting that hearing loss can be variable.²⁸

Demonstration of AFB in the ear discharge is difficult due to additional infection.¹⁹ Therefore, the clinician must maintain a high index of suspicion, perform multiple cultures and search diligently for evidence of tuberculous infection in other organs.²⁹

AFB is rarely cultured since mycobacterial counts are low in extrapulmonary tuberculosis.^{30 31} Smears of ear discharge with TOM are positive for AFB in 0–20% of cases and cultures are positive for *M tuberculosis* in 5–44% of cases.^{9–11 21} Bacteriological examination of the ear discharge is not reliable as other organisms such as *Staphylococcus*, *Pseudomonas*, *Klebsiella*, *Proteus* and *Streptococcus* can interfere with the growth of *M tuberculosis*.¹¹

The presence of secondary infection may cause delayed diagnosis. Therefore, if atypical clinical features are noted in a case of chronic otitis media, repeated cultures of the ear discharge should be obtained to rule out the possibility of TOM.

Most patients with TOM have unnecessary surgery only to have the diagnosis made from histology or tissue culture.^{9–11 22} As about 50% of patients with TOM have radiographic pulmonary tuberculosis,^{9 32} appropriate evaluation for TOM includes a chest film.

Gene amplification techniques such as PCR can be useful.³³ If the cause of suppurative infection of the middle ear is still undiagnosed, then operative biopsy may be required for diagnosis.³⁰ *Mycobacterium fortuitum* mastoiditis has also been reported after myringotomy and tympanostomy tube placement.³⁴

Anti-tuberculous therapy is the treatment of choice for TOM. The first successful treatment of TOM with antibiotics was reported in 1948 by Grief and Gould who administered streptomycin. Current standard chemotherapy uses a combination of drugs. TOM should be managed with anti-

tubercular therapy (category 1). Various chemotherapy modalities are available. The first includes a four-drug regimen in the first 2 months (isoniazid, rifampicin, pyrazinamide and ethambutol) followed by a two-drug regimen in the following 4 months (isoniazid and rifampicin). Another modality involves the use of the same four drugs (isoniazid, rifampicin, pyrazinamide and ethambutol) for a period of 6 months; this has proved effective in many centres including ours. Most patients will require at least 6 months of anti-tuberculous therapy.^{9 18 20} A notable example is a combination of pyrazinamide 500 mg, isoniazid 300 mg, rifampicin 600 mg, ethambutol 400 mg and vitamin B6 for a planned duration of 6–9 months.³⁵

Surgery is carried out for functional reconstruction, to remove bony sequester and to treat complications.^{11 21 31} Myerson and Gilbert advised radical mastoidectomy if any of the following complications develop: facial paralysis, subperiosteal abscess, labyrinthitis, mastoid tenderness or headache.³⁷ When surgery is combined with adequate chemotherapy, there is a fair chance of healing with a dry ear and a good prognosis.⁸

However, many studies have reported the use of chemotherapy alone to be sufficient. Sangeeta *et al* managed five cases with anti-tuberculous drugs with complete resolution of clinical symptoms.³⁸ Complications associated with TOM include postauricular fistula, facial nerve paralysis, labyrinthitis, tuberculous osteomyelitis of the petrous pyramid, acute mastoiditis, hearing loss and spread of the infection to the central nervous system.^{4 11 12 39}

Learning points

- ▶ Otorrhoea in a patient with known or suspected active pulmonary tuberculosis should be assumed to be tuberculous otitis media until proven otherwise.
- ▶ The clinical features in every case of chronic otitis media should be re-evaluated keeping the possibility of tuberculosis in mind.
- ▶ Early diagnosis and prompt treatment with anti-tuberculous drugs has made the condition less devastating.

Competing interests None.

Patient consent Obtained.

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