A rare case of chronic otitis externa due to *Mycobacterium tuberculosis*

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**A B S T R A C T**

Chronic otitis externa due to *Mycobacterium tuberculosis* complex is extremely rare and very few cases have been presented in the medical literature. We report here the case of an immunocompetent 68-year-old male with chronic auricular drainage, otalgia, hearing loss, external ear canal and tympanic membrane thickening for 3 years who was ultimately diagnosed with tuberculous chronic otitis externa on biopsy of external auditory canal granulation tissue using molecular diagnostic techniques. Later, sputum cultures were positive for *Mycobacterium tuberculosis* complex indicating disseminated tuberculosis. However, two plausible explanations could be pulmonary TB that disseminated to the ear canal with evidence of middle and outer otitis, or upper airway/nasopharyngeal involvement with direct extension into the middle and outer ear canals. Although extremely rare, extrapulmonary laryngeal head and neck tuberculosis should be considered in immunocompetent patients who present with chronic otitis without prior known exposure to tuberculosis when they fail standard therapy and in whom no other microbiologic cause can be identified.

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**Introduction**

Pulmonary tuberculosis is the most common manifestation of *Mycobacterium tuberculosis* infection, while extrapulmonary *Mycobacterium tuberculosis* infection is rare [1]. Excluding laryngeal forms and cervical lymphadenopathy, tuberculosis of the head and neck area makes up 2–6% of extrapulmonary tuberculosis (TB) and 0.1–1% of all forms of TB [2]. Extra-laryngeal head and neck TB has a prolonged clinical course and is difficult to diagnose, especially in cases without pulmonary involvement [2]. In a study by Prasad et al. [3], of 165 cases of TB of the head and neck region, the majority of patients (75%) did not have pulmonary involvement. We present here the case of a 68-year-old male with chronic left ear drainage subsequently diagnosed as chronic otitis externa due to *M. tuberculosis* complex.

**Case presentation**

A 68-year-old male was referred to Otorhinolaryngology at Mayo Clinic in July of 2014 for persistent left ear drainage that began approximately 3 years ago. He was otherwise healthy and without any constitutional symptoms. In 2013, a tympanoplasty was performed due to persistent drainage, followed by a revision tympanomastoidectomy in January of 2014. Unfortunately, his hearing did not improve and the drainage continued. Two separate bacterial cultures that were obtained had no growth. He received ear drops ciprofloxacin 0.3%/dexamethasone 0.1% and beta-dine without resolution of his symptoms.

On his presentation in 2014, his ear examination revealed mild myringitis, posterior ear canal thickening, and drainage. The tympanic membrane was intact. The diagnosis of external auditory canal atopic dermatitis was initially entertained and he was prescribed Tacrolimus–neomycin and polymyxin B sulfates and hydrocortisone otic solution otic suspension. He had moderate improvement in symptoms approximately one month later. Attempts to wean him off daily Tacrolimus–neomycin and polymyxin B sulfates and hydrocortisone otic solution otic suspension resulted in...
acute exacerbations of mild pain and ear drainage. Laser myringoplasty for chronic myringitis was recommended. In October 2014, laser myringoplasty on the left side was performed, but the patient had developed severe external auditory canal granulation tissue that was worrisome for squamous cell carcinoma, and consequently, multiple deep biopsies of the external canal were obtained along with debulking of granulation tissue. Hematoxylin and eosin (H&E) stained sections showed reactive squamous epithelium with necrotizing granulomatous inflammation (Fig 1). Acid-fast bacilli (AFB) stain was positive for many acid-fast bacilli (Fig 2) while Grocott’s methenamine silver stain was negative for fungal organisms. There was no evidence of dysplasia or squamous cell carcinoma. Real-time PCR of the formalin-fixed paraffin embedded tissue was positive for M. tuberculosis complex.

Additionally, an acid-fast stain performed on a swab collected from the left anterior ear canal was negative, but grew acid-fast bacilli at 13 days in Middlebrook broth cultures (BACTEC MGIT 960, Becton Dickinson, Sparks, MD) that was identified by nucleic acid hybridization probe (AccuProbes, Hologic GenProbe, Bedford, MA) as M. tuberculosis complex. Real-time PCR to determine the species within the M. tuberculosis complex was performed as previously described [4] and identified the growth from the 7H11 biplate subculture as M. tuberculosis. Culture of the swab on solid media (7H11) biplate grew dry, rough, nonchromogenic colonies after 15 days that were acid-fast with cording by Kinyoun stain. Matrix-assisted laser desorption/ionization time of flight mass spectrometry (MALDI-TOF MS) using the Bruker MALDI Biotyper (Bruker Daltonic, Billerica, MA) and version 3.1 software and mycobacteria database 2.0 identified the isolate as M. tuberculosis complex with a score of 2.193. Furthermore, two additional surgical specimens, 1 swab from the left ear and tissue from the biopsy of the left ear were culture positive for M. tuberculosis complex.

The finding of M. tuberculosis from the ear specimens was surprising and the patient was then evaluated by an Infectious Diseases specialist. The patient denied any cough, fever, weight loss, or hemoptysis. He lived most of his life in the Midwest with travel only to Florida in the winter and without any travel outside of the continental United States. He is a retired teacher. He denied any known TB exposure or history of incarceration. He does not recall having testing for latent tuberculosis (i.e. purified protein derivative [PPD] or gamma interferon release assay testing) in the past. His laboratory findings including a complete blood count, liver function testing and creatinine were normal. HIV serology testing was negative. QuantiFERON-TB Gold In-tube testing was positive (tuberculosis antigen value 11.97 IU/mL). His chest x-ray showed no abnormalities. Three induced sputa performed at an outside hospital were negative by acid fast smear. A separate induced sputum collected at our facility was negative by acid-fast smear, but mycobacterial culture grew two colonies of M. tuberculosis after 36 days. A CT scan of the head from a different institution performed on November 2013 and June 2014, were reviewed by our radiologist and revealed diffuse narrowing of the left external auditory canal, opacification of the left middle ear and mastoid air cells, and destructive changes involving lateral left mastoid air cells (Fig 3). The mastoid area had been previously debrided elsewhere and involvement at the time of presentation was thought to be limited to the soft tissue of the external auditory canal. Treatment for extrapulmonary TB was initiated with rifampin 600 mg daily, isoniazid 300 mg daily, pyrazinamide 2000 mg daily, and ethambutol 1600 mg daily for 2 months. After 2 months, the ethambutol and pyrazinamide were discontinued, as the isolate was pan-sensitive. The plan was to treat with a 12 month
course of isoniazid and rifampin. This is not the standard length of treatment—which would be 6 months. We extended the duration to 12 months because his course was complicated with recurrent otorrhea after initial improvement on therapy. We wanted to rule out recurrent TB or identify an alternative explanation. Repeated biopsy of the external ear canal and multiple subsequent mycobacterial cultures were performed and were negative for M. tuberculosis, however they grew Stenotrophomonas. This was thought to be a secondary infection of the external ear canal requiring the addition of oral sulfamethoxazole/trimethoprim and ciprofloxacin 0.3%/dexamethasone 0.1% eardrops to his antimicrobial regimen. His otorrhea eventually resolved and his ENT exam normalized.

Discussion

Diagnosis of chronic otitis externa due to TB is seldom considered in an immunocompetent adult host without pulmonary involvement. Moreover, in reviewing the medical literature we found that chronic otitis externa due to M. tuberculosis is exceedingly rare, and this is also true for non-tuberculosis mycobacteria. Several case reports and reviews discuss TB as a cause of chronic suppurative otitis media and mastoiditis [5–7]. Most of these patients present with chronic serous ear drainage, otalgia, often unilateral hearing loss and facial palsy [7,8]. Our patient had recurrent otitis media and auricular drainage for approximately 2 years with several surgeries, bacterial cultures, and antibiotic treatment without improvement. Atopic dermatitis was initially considered and treated with tacrolimus–neomycin and polymyxin B sulfates and hydrocortisone ointment solution otic suspension with some improvement, but any attempts to wean off therapy resulted in acute exacerbations. Given this patient’s unremarkable chest x-ray, sputum positive for TB, the most likely pathogenesis began with TB present in the upper airways/nasopharynx with extension into the middle ear via the Eustachian tube, and progressing into the external ear canal. A similar case was reported in the Netherlands of a man with bilateral otitis media, progressive hearing loss, and tinnitus. [9] Fiber endoscopy a mass with purulence was seen in the left middle nasal passage extending up to the left Eustachian tube.

[9] A nasal endoscopy was not performed in our patient, but may have been of helpful in discriminating between pulmonary TB that disseminated to the ear canal with evidence of middle and outer otitis, or upper airway/nasopharyngeal involvement with direct extension into the middle and outer ear canals.

After the diagnosis of disseminated tuberculosis was made, it was later discovered that the patient’s wife had a positive quantiFERON-TB testing, and was treated for latent tuberculosis infection.

Conclusion

Tuberculous chronic otitis externa presents a rare diagnostic challenge that requires a multidisciplinary approach—infec tious diseases, otolaryngology, and clinical microbiology for prompt diagnosis. This diagnosis should be considered in a patient who progresses on standard therapy, even if immunocompetent, and who has no other microbiological cause identified.

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