Unusual presentation of tubercular mastoid cyst

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Abstract
Tuberculosis in the otomastoid compartment is extremely rare. The classical presentation of tuberculous otitis media includes chronic otorrhea, severe hearing loss irrespective of the disease, multiple perforations, and pale granulations in the middle ear cavity. However, most present with non-specific signs and symptoms making it a diagnostic dilemma. Here, we report a case that presented with a bony mastoid cyst which is the first case report to the best of our knowledge. A 12-year-old boy visited our center with a complaint of chronic ear discharge and hearing loss not improving with conventional antibiotic therapy. The computed tomography scan of the temporal bone revealed soft tissue opacification in the mastoid along with features suggestive of cholesteatomatous chronic otitis media. The patient underwent mastoidectomy under general anesthesia, which revealed a bony mastoid cyst filled with turbid fluid along with granulation tissues in the middle ear cleft without cholesteatoma. Final histopathology revealed tuberculous otitis media. Antitubercular chemotherapy was started and the patient's condition recovered. Primary tuberculous mastoiditis is a rare clinical entity that requires a high index of suspicion. It can also present as chronic otorrhea with mastoid cyst, and thus need to be considered as one of the differential diagnoses.

Keywords
Tuberculosis, otitis media, mastoid cyst

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Introduction
Tuberculosis (TB) is one of the major infectious and communicable diseases and falls under the top 10 causes of death worldwide.¹ The causative organism is Mycobacterium tuberculosis bacteria which mainly affects the pulmonary system; however, in 15%–30% of cases, the infection can occur extrapulmonary.² In 2019, 16% of the 7.1 million incident cases were extra-pulmonary tuberculosis (EPTB).¹ Among the EPTB, tuberculous otitis media (TOM) is one of the rarest and accounts for 0.05%–0.9% of chronic otomastoiditis.³,⁴ The incidence of TOM had decreased enormously since the commencing of the 21st century, but there has been a reemerging of the infection due to HIV infection.³,⁵,⁶ TOM is more common in children and the classical presentation is painless foul-smelling otorrhea with severe mixed hearing loss which do not match with the severity of the disease, along with pale granulations in the middle ear cavity and multiple tympanic membrane perforations.⁷ However, most cases present with non-specific clinical features making it a diagnostic challenge.⁸ In the present case, we report an atypical presentation of TOM which presented with chronic ear discharge along with the bony mastoid cyst. No literature has reported TOM presented as a mastoid cyst and to the best of our literature review, this would be the first case report. Thus, we aim to expand the knowledge on the presentation of TOM and help the surgeon to consider the differential diagnosis.

Case summary
A 12-year-old boy presented with intermittent, foul-smelling, occasionally blood-stained left ear discharge for 3 years which did not improve with conventional antibiotic therapy. He had no other features suggestive of TB such as cervical lymphadenopathy, weight loss, and night sweats. Otoendoscopic examination

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revealed a fleshy polyp at the posterior superior and attic area associated with thick yellowish discharge. The pars tensa of the tympanic membrane was intact (Figure 1). Computed tomography (CT) temporal bone revealed soft tissue opacity in the middle ear with the ossicular erosions and destructions. There was also soft tissue density in the mastoid region with loss of septation and dehiscence of tegmen and sigmoid plate. However, the facial nerve canal, internal acoustic meatus, cochlea, and vestibule were intact (Figure 2). The audiometric examination revealed severe mixed hearing loss. The patient underwent modified radical mastoidectomy under general anesthesia, via a postaural approach using the out-to-in technique (Figure 3(a)). Upon drilling the mastoid, a cystic lesion was encountered (Figure 3(b)). The drilling was done around the cyst; however, it got punctured during the procedure and yielded dirty turbid whitish fluid. The remaining cyst and tissues were removed and sent for histopathological evaluation. There was a sequestrum as well which was also removed. The granulation tissues at the attic were removed. Posterior bony canal wall was drilled out and canal wall down mastoidectomy was performed. The lenticular and long process of incus was necrosed and removed. The malleus head was removed to evaluate the anterior epitympanum (attic). Stapes suprastructure was partially necrosed (Figure 3(c)). Temporalis fascia graft was placed after clearing the disease. There were no postoperative issues. The cyst wall along with granulations tissues were sent for histopathological examinations (Figure 3(d)). The histopathological report showed epithelioid cell granulomas, Langerhan’s type giant cell, chronic inflammatory cells, and granulation tissue, consistent with TB. The cyst wall lining epithelium was attenuated with chronic inflammatory cells infiltration and giant cell reaction (Figure 3(e)). The antitubercular medications were started for the patient, and finally, the patient recovered after completion of chemotherapy (Figure 3(f)). The patient was followed up for 1 year, the cavity healed however, the hearing did not improve.

Discussion

TB infection in the middle ear cavity, mastoid, and temporal bone is extremely rare. The first case of TOM was reported in 1853.7 The classical presentation is usually absent and most cases have non-specific presentations making it a diagnostic challenge.9 Epidemiologically and clinically, significant changes have been observed in the tubercular otomastoiditis.
after Bacillus Calmette-Guérin (BCG) vaccination and antitubercular chemotherapy. TOM has been considered to affect more in the younger children and is more in low-income countries.

The pathophysiology of TOM is unclear and controversial. The bacterium can enter the middle ear via a hematogenous route, or through mucus aspiration via the Eustachian tube, or can be directly implanted through the external auditory meatus and tympanic membrane perforation. Clinically patients present with painless otorrhea not responding to treatment, disproportionately huge sensorineural hearing loss, multiple perforations, and pale granulation tissues. In 90% of the cases, patients suffer from severe hearing loss. The hearing losses most often happen early on and are severe, which can be sensorineural, and mixed or conductive. Sensorineural hearing loss can be due to vasculitis of the cochlear veins, deposition of the immunocomplex in the cochlea, or due to the presence of granulations affecting the acoustic nerve. Persistent and pronounced dilatation of blood vessels in the anterior tympanic membrane for a long time is considered pathognomonic of TB of the middle ear.

However, most patients do not present with these classic signs and symptoms. Hence, the diagnosis is not so easy and is usually recognized late, mainly due to a low index of suspicion, its rare prevalence, and non-specific clinical presentation mimicking chronic otitis media refractory to standard antibiotics. Complications of TOM include facial nerve palsy, ossicular necrosis, destruction of the mastoid and petrous bone, periauricular fistulas, lymphadenopathy, and intracranial tuberculomas or abscesses. Nevertheless, early diagnosis and treatment play a vital role to avoid the spread of the disease and permanent injury to hearing and facial nerve function. In our case, we had atypical presentation

Figure 3. (a) Postaural incision, out-to-in approach. (b) Mastoid cyst occupying the mastoid cavity. (c) Final view after canal wall down mastoidectomy. (d) Gross picture of cyst wall. (e) Histopathological finding of the granulation and cyst wall. Attenuated cyst wall lining (**) and multinucleated giant cell (solid black arrow) and (f) postoperative picture after completion of antitubercular chemotherapy.
TOM as a mastoid cyst. Some of the rare presentations include Bezold’s abscess and zygomatic abscess.7,9

A CT scan on the mastoid revealed that TOM is associated with less sclerotic changes of the temporal bone, while cortical bone destruction was a more frequent finding.6 The finding of protuberant granulation tissue in the mastoid, bony sequestrum, or fallopian canal dehiscence should alarm the surgeon about the possibility of tuberculous mastoiditis.11 Because of the poor specificity of the temporal bone imaging, the gold standard for TOM diagnosis remains microscopic evaluation, bacterial culture, drug sensitivity testing, and histopathological examination of specimens obtained from the middle ear, as recommended by the European guidelines.11

Treatment of TOM is chemotherapy with antitubercular therapy.2,6 But opinion varies its duration of treatment. Some regard it as part of bone TB so they are advised for a longer treatment of 9–12 months.9,10 Some regards it as EPTB, so treatment as EPTB for 6 months.5 We started antitubercular medications as per our national guidelines that follow the guidelines of WHO. The regimen includes the first 2 months of the intensive phase of HRZE (Isoniazid, Rifampicin, Pyrazinamide, and Ethambutol) followed by 4 months of the continuation phase of HR (Isoniazid and Rifampicin).

The surgical management is mandatory in two cases: first is for a diagnostic purpose when the clinical suspicion is highly likely, but polymerase chain reaction (PCR) analysis and bacterial culture of ear discharge fail to identify the bacteria. The second surgical indication is to complete treatment when chemotherapy is not sufficient, to eliminate the infectious process from the middle ear cleft and the mastoid compartment.11 At present, the surgical treatment needs to be held in reserve for cases with complications including subperiosteal abscess, facial palsy, and sequestrum removal.9,10

**Conclusion**

Primary tuberculous mastoiditis is a rare clinical entity. Most cases present with non-specific signs and symptoms, and thus require a high index of suspicion. It can also present as chronic otorrhea with mastoid cyst which should be considered as one of the differential diagnoses.

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**Ethical approval**

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**Informed consent**

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