

## CASE REPORT

### Tuberculous Otitis Media-Diagnosis and Treatment of Four Cases

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We present four cases of tuberculous otitis media that were diagnosed and treated at our clinic during a five-year period. Clinical manifestations, required additional diagnostic techniques, treatment, complications and diagnostic pitfalls of tuberculous otitis media are presented and discussed along with published literature. Because tuberculosis is such an uncommon cause of chronic infection in the middle ear and mastoid the index of suspicion is low and the diagnosis is quite difficult. In order to avoid delayed diagnosis and irreversible complications, it is always necessary to think about tuberculosis in cases that present with chronic painless otitis media, perforated tympanic membrane and facial nerve palsy.

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#### Introduction

Tuberculosis (TB) has a long history. It is presumed that it may have killed more persons than any other infectious disease so far<sup>[1]</sup>. TB accounts for 2.5 % of the global burden of disease and it currently holds the seventh place in the global ranking of causes of death. Unless intensive efforts are made, it is likely to maintain that position through to 2020, despite a substantial projected decline in disease burden from other infectious diseases<sup>[2,3]</sup>. The majority of the extrapulmonary forms of TB affect organs with suboptimal conditions for bacillary growth. For this reason, the extrapulmonary disease generally has an insidious presentation, a slow evolution and paucibacillary lesions and/or fluids. Access to the lesions through secretions and body fluids is not always possible, and for this reason, invasive techniques may be necessary in order to obtain material for diagnostic investigation. Tissues and/or

body fluids should be submitted to laboratory examination, in particular bacteriological culture for Mycobacteria and histopathological analysis. In immunocompetent patients, the extrapulmonary forms only occasionally coexist with active pulmonary TB. Nevertheless, the chest X-ray is mandatory for the evaluation of evidence of primary infection lesions. The most common extrapulmonary site is pleural TB, and one of the rarest is middle ear. Tuberculous otitis media (TOM) accounts for between 0.05% and 0.9% of chronic middle ear infections<sup>[4,5,6,7]</sup>. Because TB is such an uncommon cause of chronic infection in the middle ear and mastoid the index of suspicion is low and the diagnosis is quite difficult. Many physicians are unfamiliar with its presenting features and laboratory studies necessary for correct diagnosis. Clinical signs are often variable and different from the classical description. False-negative cultures of Mycobacteria often occur because other interfering

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bacteria are present in the specimen<sup>[7]</sup>. Generally, middle ear TB is unilateral, presenting with painless otorrhea, multiple small perforations of the tympanic membrane and concomitant peripheral facial nerve palsy and bone necrosis<sup>[5]</sup>. Delayed diagnosis leads to delayed appropriate treatment thus followed by increased risk of irreversible complications such as permanent hearing loss or facial nerve palsy. TOM is successfully treated with combined antituberculous medication, for at least 6 months, but the role of surgery in treating TOM is very important<sup>[5,7,8]</sup>. We present four cases that were diagnosed and treated at our clinic during a five-year period with the idea of reporting clinical manifestations and complications of TOM.

## **Case presentations**

### **Case 1**

A 75-year old male patient was admitted to our clinic in January 2006, with right-sided peripheral facial paralysis that developed 3 days before admission (Grade 2 according to House-Brackmann Scale). Patient also complained about chronic aural discharge (he had a ten year history of right side otorrhea that was refractory to medical treatment) and unilateral hearing loss that intensified during time. Clinical examination revealed total perforations of the right tympanic membrane. Pure tone audiometry showed deafness in the right ear and a moderate to severe sensorineural hearing loss in the left ear. Initial laboratory data and chest X-ray were normal, and culture of ear drainage was negative. Computerised tomography (CT) of the temporal bones showed features of right-side mastoiditis with soft tissue attenuations in middle ear. On the second day of hospitalisation, patient underwent right radical mastoidectomy with decompression of the tympanic part of the facial nerve. During operation, we noted that the mastoid, attic and middle ear cleft were encompassed in granulation tissue and the ossicles were eroded. After decompression of the facial nerve, signs of neural edema could be observed. We also noted a fistular structure in the lateral semicircular channel.

Histopathological examination of the granulations obtained during surgery revealed presence of numerous TB granulomas with caseous necrosis. Ziehl-Neelsen stain for acid-fast bacilli was negative. Patient started

with antituberculous medications (isoniazid, rifampicin, ethambutol and pyrazinamide) and physical therapy for facial nerve function improvement. During a 12 month follow-up patient recovered and facial nerve function significantly improved.

### **Case 2**

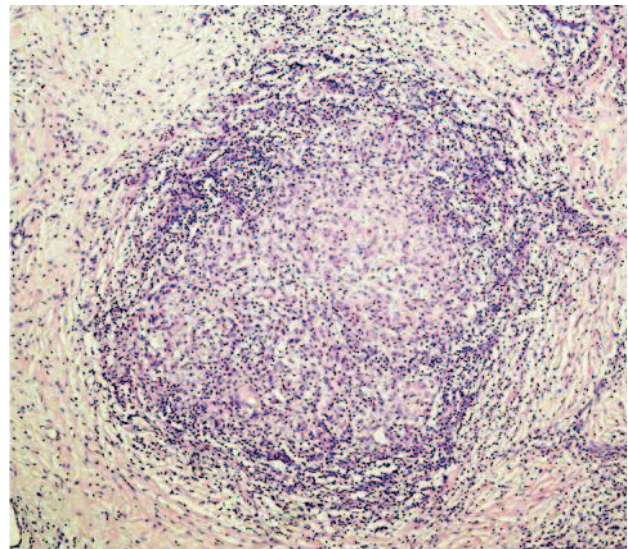
A 56 years old female patient was admitted to our clinic with a diagnosis of chronic otitis after she was unsuccessfully treated with antibiotics for several months in another medical facility. Patient had a long history of left-side otorrhea and total unilateral conductive hearing loss along with occasional vertigo and headaches. Peripheral facial palsy ensued (Grade 4 according to House-Brackmann Scale), with general deterioration and various psychological symptoms. Clinical examination revealed granulations and purulent discharge present in obliterated external auditory canal. CT of temporal bones was ordered in first 24 hours of hospitalisation and it showed features of soft tissue attenuations in external auditory canal and middle ear along with destruction of ossicular structures (Figure 1). On the second day of hospitalisation, patient underwent radical mastoidectomy. During surgical intervention, granulations, numerous bone sequestrations and destruction of ossicular bones were noted. Histopathology revealed numerous tuberculous granulomas, consistent with diagnosis of middle ear TB (Figure 2). Further analysis showed border-line values of purified protein derivate test, and chest X-ray was normal. During follow-up and therapy with antituberculous medications (isoniazid, rifampicin, ethambutol and pyrazinamide), facial nerve function improved significantly after 6 months.

### **Case 3**

A 42 years old male patient was admitted to our clinic with a diagnosis of chronic otitis after he was unsuccessfully treated with antibiotics for several months in another medical facility. Patient had a long history of right-side painless otorrhea and unilateral mixed hearing loss. He underwent surgery and, during the first step of tympanoplasty in middle ear cavity, presence of profuse, white inflammatory debris, along with signs of bone sequestration was noted. Chest X-ray ensued in next 24 hours of hospitalisation and it revealed a miliary



**Figure 1.** Tuberculous otitis media complicated by destruction of middle ear bony structures including ossicular bones and facial canal (rectangle). Computerised tomography scan of temporal bones, axial view.

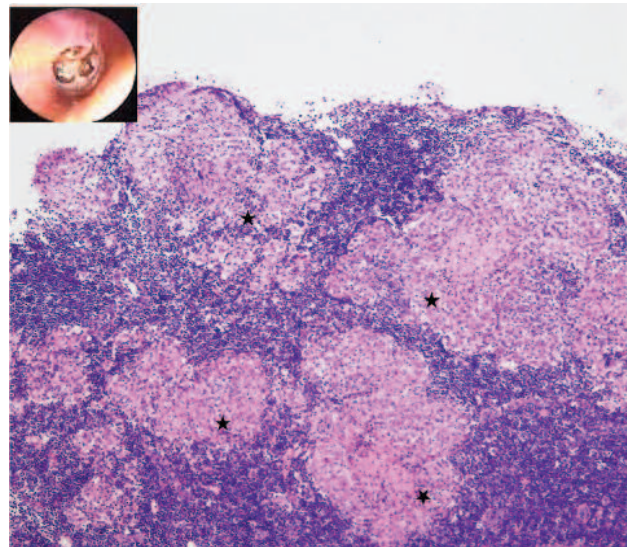


**Figure 2.** Tuberculous otitis media: Tuberculous granuloma from a middle ear biopsy, Hematoxyllin & Eosin, original magnification x 400.

TB. Patient successfully recovered after receiving 4 combined antituberculous medications, during a period of 6 months.

#### Case 4

A young female patient (35 years) was treated for several years as unilateral, right tubo-tympanic otitis and allergic rhinitis in another medical facility. There were occasional episodes of otorrhoea, mixed hearing loss, vertigo and tinnitus. Patient also had difficulties while breathing through the nose. At admission, clinical examination revealed two minor perforations (Figure 3) at upper and lower quadrant of tympanic membrane („moth-eaten“ appearance). Otomicroscopical examination that ensued suggested some form of adhesive otitis, with signs of hyalinisation of middle ear mucosa. During surgery, epipharyngoscopy was also performed because adenoid-like vegetations were present in epipharynx. CT of temporal bones was consistent with features of chronic otitis. Histopathologically, vegetations removed from epipharynx were infiltrated with TB granulomas (Figure 3.). Chest X-ray was normal and there were no signs of TB in other organs. Patient successfully recovered after receiving antituberculous medications during a 6-month period.



**Figure 3.** Perforated, „moth-eaten“ tympanic membrane (upper left quadrant) and biopsy taken from epipharyngeal mucosa vegetations showing numerous tuberculous granulomas (asterisks). Hematoxyllin & Eosin, original magnification x 200.

#### Discussion

The involvement of the temporal bone by TB was first described by Jean Louis Petit in 18<sup>th</sup> century<sup>[9]</sup>. Pathogenesis of TOM is related to three mechanisms-direct spread from the Eustachian tube, hematogenous spread from another TB focus and direct implantation through tympanic membrane perforation. About 50% of patients with otologic evidence of TB have



radiographic pulmonary disease. We presented 2 patients with TOM that were without evidence of concomitant pulmonary disease. One of our patients had miliary TB, and one patient had epipharyngeal TB. TOM has high prevalence among children and occurs more frequently in men<sup>[6,8,9-12]</sup>. In the series published by Cho et al. 30 of 52 patients were female and only 5 were younger than 15 years of age, and the highest incidence was reported among patients in their 30s<sup>[7]</sup>. Our patients ranged from 35 to 56 years (mean age 44 years), 2 of them were female and one was male. The duration of ear symptoms before presentation is reported to vary substantially, from less than 1 month up to 40 years<sup>[4-12,14,15]</sup>. All our patients were treated unsuccessfully with antibiotics during a period of several months up to several years. Painless otorrhea is the most consistent finding, but in many cases otalgia was reported<sup>[7]</sup>. Some patients report dizziness, some tinnitus and some present with conductive or mixed hearing loss and peripheral facial palsy (in range from 9.6% up to 45% of reported cases) and tympanic membrane perforation and/or adhesions<sup>[4-8,14,15]</sup>. Two of our patients had mixed hearing loss and one had a conductive hearing loss. One patient had peripheral facial palsy, and one had two small tympanic membrane perforations that had „moth-eaten“ appearance. The patient with a peripheral facial palsy had the shortest duration of symptoms (they were present for several months), which is consistent with reports of Cho et al<sup>[7]</sup>. There was about 20% of reported cases that had some form of bony destruction, usually in the external auditory canal, mastoid bone or ossicular bones. Temporal bone CT usually reveals soft tissue attenuations that fill the mastoid and external auditory canal. Such CT findings were also present in two of our cases. High rate of missed preoperative diagnoses may be low index of suspicion, low incidence of radiological evidences of pulmonary TB and atypical clinical presentation. Concomitant pulmonary TB should suggest etiology in a patient with chronic otitis media; however, TOM should not be excluded when otorrhea occurs in a patient without pulmonary disease<sup>[8]</sup>. Most of the reported cases that had no evidence of pulmonary disease were diagnosed from obtained surgical specimens, either by histopathology or by tissue cultures<sup>[4-12]</sup>. Two of our cases were diagnosed by histopathology

and one by chest X ray that showed miliary TB in lungs. In a study by Cho et al. patients who had surgery and chemotherapy achieved recovery much earlier than patients that had only chemotherapy<sup>[7]</sup>. The role of surgery is important in removing bone sequestrations and granulation tissue along with tympanoplasty, but also for improvement of ear drainage. Surgery provides prompt and accurate diagnosis and, along with chemotherapy, complete healing as well<sup>[7,9,13]</sup>. All our patients were treated surgically, and during follow-up combined antituberculous chemotherapy ensued, which secured their recovery.

Cases that we presented had a variety of diagnostic pitfalls, with chronic otitis as a dominant clinical feature. Otogenic facial nerve palsy (which was present in two of our cases) is most commonly connected with a non-specific inflammatory process, and is routinely treated like one. Additional diagnostic procedures should be applied in cases that present with even the slightest suspicion about specific or any other pathological process. Unilateral hearing loss, especially if connected with other discrete clinical signs such as mild otorrhea and intact or „moth-eaten“ tympanic membrane can remain without accurate diagnosis for a long period of time. In those cases one should always consider otological disease in the context of loco-regional pathology (paranasal or pharyngeal). Pathological process located in epipharyngeal region can remain without symptoms, with domination of otological symptomatology. In case number 4, during a diagnostic procedure in a patient that had mostly otological symptomatology, a granulomatous epipharyngeal inflammation was detected and thus successfully treated. Epipharyngeal TB is exceedingly rare and can be easily clinically overlooked leaving the patient without the accurate diagnosis and treatment. Each time when there is a clinical suspicion of pathological process located in epipharyngeal region, we suggest direct endoscopy and biopsy as an accurate diagnostic tool.

Clinical presentation of TOM in cases with concomitant miliary TB can range from discrete symptoms up to bony destruction; additional diagnostic is required when the condition of the patient doesn't improve after successful surgery. Some of recently published papers

suggest Quantiferon-TB Gold Test as a very useful diagnostic tool for diagnosing latent or active TB [16-18].

In summary, we presented diagnostic pitfalls, complications and additional diagnostic techniques required during the treatment of patients with discrete symptomatology of TOM.

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