

**Case Report****Tuberculous Otitis Media: a case report of hearing impairment in developing country****Giovanni Reynaldo\*, Bernadina Chyntia Carsantiningrum\*, Harim Priyono\*\***

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Ciptomangukusumo Hospital, Jakarta**ABSTRACT**

**Background:** Tuberculosis is one of major health problems in developing countries, especially extrapulmonary tuberculosis. Tuberculous otitis media (TOM) is one of extrapulmonary manifestations which is a rare phenomenon characterized by painless otorrhea, insidious onset of ear discharge, multiple perforations in the tympanic membrane, and pale granulation tissues in middle ear cleft. **Purpose:** Reporting one rare case of TOM. **Case Report:** A 58-year-old male came with painless otorrhea and recurrent hearing impairment. Tympanomastoidectomy was carried out to repair the tympanic membrane, to cleanse the secret from the middle ear, and to obtain sample for biopsy. Histopathological examination showed necrotizing granuloma which contained mycobacterium tuberculosis infection. Medical treatment was administration of anti tuberculosis drugs. **Clinical Question:** How to establish TOM diagnosis? **Review Method:** Searching for literature evidence through Google Scholar. **Result:** The search obtained 20 journals which in accordance with the inclusion and exclusion criteria. There were similarities on clinical and therapeutic symptoms with this reported case. **Discussion:** In the reported case, the probable pathophysiology was bacterial aspiration through the Eustachian tube, which was just diagnosed during pre-operative screening. There was no apparent pulmonary tuberculosis symptom. Diagnosis TOM with mastoiditis was difficult, it required high skilled accuracy. **Conclusion:** TOM is a rare manifestation of extrapulmonary tuberculosis. High suspicion of TOM is needed in patients who did not respond to standard treatment. Treatment includes administration of anti-tubercular drugs, and surgical procedure to cleanse the secretion and granulation tissues. Permanent hearing loss can occur in cases of delayed diagnosis.

**Keywords:** Otitis media, tuberculous, hearing loss, otorrhea**ABSTRAK**

**Latar belakang:** Tuberkulosis merupakan masalah kesehatan di negara berkembang, terutama tuberkulosis ekstra pulmonal. Otitis media tuberkulosa (OMT) merupakan salah satu manifestasi tuberkulosis ekstra pulmonal yang jarang terjadi dan ditandai dengan gejala klinis berupa keluar cairan dari telinga tanpa disertai rasa nyeri, onset penyakit berjalan lambat, terdapat perforasi multipel pada membran timpani, dan jaringan granulasi pucat di rongga telinga tengah. **Tujuan:** Melaporkan satu kasus OMT yang jarang ditemukan. **Laporan Kasus:** Seorang laki-laki 58 tahun datang dengan keluhan keluar cairan dari telinga tanpa disertai rasa nyeri, dan ada gangguan pendengaran berulang. Pada pasien dilakukan tindakan bedah timpanomastoidektomi untuk memperbaiki membran timpani yang rusak, membersihkan sekret, dan melakukan biopsi jaringan. Hasil pemeriksaan histopatologi didapati jaringan granuloma nekrotikans yang menunjukkan adanya infeksi *Mycobacterium tuberculosis*. Diberikan terapi medikamentosa obat anti tuberkulosa. **Pertanyaan Klinis:** Bagaimana menegakkan diagnosa OMT? **Telaah literatur:** Penelusuran bukti kepustakaan melalui Google Scholar. **Hasil:** Penelusuran menghasilkan 20 jurnal yang memenuhi kriteria inklusi dan eksklusi dan memiliki kesamaan gejala klinis dan terapi dengan kasus yang dilaporkan. **Diskusi:** Pada kasus ini kemungkinan patofisiologinya adalah aspirasi bakteri melalui tuba eustachius, dan OMT baru terdiagnosis saat dilakukan skrining pra-operasi. Tidak didapati gejala tuberkulosis paru. Diagnosis OMT dengan mastoiditis cukup sulit, diperlukan

ketelitian yang tinggi. **Kesimpulan:** OMT merupakan manifestasi tuberkulosis ekstra pulmonal yang jarang terjadi. Perlu kecurigaan yang tinggi adanya OMT pada pasien yang tidak responsif terhadap pengobatan standar. Penatalaksanaan meliputi pemberian obat anti tuberkulosa, dan tindakan bedah untuk mengeluarkan dan membersihkan sekret dan jaringan granulasi. Gangguan pendengaran permanen bisa terjadi pada kasus yang penatalaksananya terlambat.

*Kata kunci:* Otitis media, tuberkulosa, gangguan pendengaran, otorrhea

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## INTRODUCTION

Tuberculosis (TB) has high prevalence rate particularly in developing countries. WHO estimated 8 million reported TB cases per year worldwide.<sup>1,2</sup> TB incidence is 55% in Asia and 31% in Africa.<sup>1</sup> A third of the world's population is affected by TB, and that 9 million new cases and 3 million deaths are recorded every year because of it.<sup>3</sup> *Mycobacterium tuberculosis* is the most frequently responsible bacillus of TB in humans (95%), although other bacilli of the *Mycobacterium tuberculosis* complex (*bovis*, *africanum*, *microti*) have also been reported as causative agents.<sup>4</sup>

Pulmonary TB was found in 85% cases, while 15% was extrapulmonary TB.<sup>2,3,4</sup> The most often found extrapulmonary TB was lymph glands (48.9%), pleura (25.5%), bone (22.7%), urinary tract (5.7%), and meninges (5%).<sup>2</sup> The highest incidence of extrapulmonary TB in head and neck is cervical lymphadenitis (95%). It could also affect middle ear area, nasal cavity, oropharynx, nasopharynx, parotid and submandibular glands, esophagus, palatum, tongue, trachea, larynx, and oral mucosa, with prevalence rate around 1%.<sup>3</sup>

TB of the ear is a rare entity and in most cases, the clinical features resemble that of chronic otitis media. The diagnosis is often delayed due to varied clinical presentations and this can lead to irreversible

complications.<sup>2</sup> Tuberculous otitis media (TOM) as a rare case is 0.05-0.9% from the whole chronic infections in the middle ear.<sup>4,5,6</sup> The incidence had decreased compared to the incidence rate at the beginning of 20<sup>th</sup> century, as high as 1.3% - 18.6%.<sup>3</sup> It could probably be the result of the environment improvement, better laboratory facilities, BCG vaccine, and antitubercular drugs vast availability.<sup>3,6</sup> A study by Palma et al.<sup>7</sup> revealed that adult mastoiditis was estimated around 0.99 case per year per 100.000 population. Up to date, the diagnosis of TOM remains a significant challenge for otolaryngologists due to unspecific clinical sign and symptoms. Therefore, high suspicion of TOM is required in patients who did not respond to standard treatment of chronic middle ear infection.<sup>4</sup>

The gold standard for establishing TOM diagnosis is microbiological culture and histopathological examination (HPE) of the tissue obtained from the middle ear and mastoid. If the presence of *Mycobacterium* is proven, the standard antitubercular treatment (ATT) should be administered.<sup>5</sup>

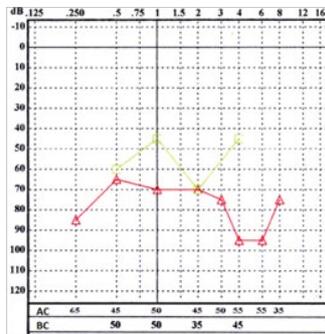
The purpose of presenting this case was to report one rare case of tuberculous otitis media in order to increase the alertness of colleagues regarding TOM's clinical symptoms and promoting early diagnosis to avoid otological permanent damage.

**CASE REPORT**

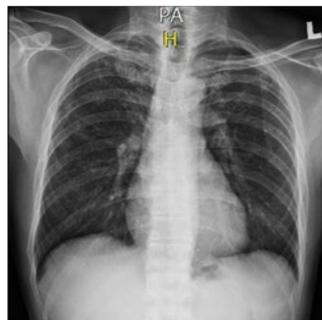
A 58 years old male came with hearing decrease in his right ear for one month. There was no flowing discharge from the ear when the complaint was first noticed. The patient had a history of pulmonary TB for 20 years and had completed the TB therapy. There were no weight loss, fever, coughing, sputum production, nor night sweats. No history of smoking, alcohol consumption, or other comorbid. No history of right ear trauma. Upon physical examination, patient looked healthy, not pale, vital signs were good, no

fever. Lung and heart examination found no rhonchi nor other abnormalities. He was given standard antibiotic and supportive therapy for otitis media with effusion for 2 weeks, with no significant result.

On his next visit, otoscopy examination of the right ear revealed pale, opaqued, intact tympanic membrane befitting otitis media with effusion appearance. Rinne test showed a conductive deafness. Audiometry response of the right ear was 70-100 dB and the left ear was normal under 25 dB (Figure 1).



**Figure 1.** Pre-operative audiometry of right ear



**Figure 2.** X-ray thorax pre-mastoidectomy and tympanoplasty, image of active lung tuberculosis



**Figure 3.** CT-Scan mastoid postoperative

It was decided to install a ventilation tube on his right ear. During the insertion of ventilation tube, a purulent discharge was flowing out of the right middle ear. Hearing improvement was gained after that otologic measure. One month afterwards, the patient came to the clinic with the same complaint as prior to the installation of ventilation tube, this time also accompanied with tinnitus. Otoscopy examination revealed symptoms of right ear otitis media with effusion. Nasoendoscopy examination found no tumor nor blockage of the Eustachian tube. Again he was given antibiotic and symptomatic medication with no positive result. On the next visit, it was discovered a perforation of the right ear tympanic membrane with otorrhea. It was planned to perform tympanomastoidectomy surgery, as to repair the tympanic membrane, to cleanse the secret from middle ear cavity, and to obtain a biopsy for histopathological examination.

Histopathological examination of the middle ear tissue showed a tuberculous specific granulomatous inflammation process. The pre-operative thorax X-ray clearly showed image of pulmonary TB (Picture 2). Diagnosis of OMT and mastoiditis were confirmed, patient was advised to take anti tubercular treatment (ATT) consisted of rifampicin 600 mg, isoniazid 300 mg, pyrazinamid 1000 mg, ethambutol 1000 mg, and curcuma 1 tablet per day.

Evaluation was performed after 9 months ATT. The thorax CT scan still showed pulmonary TB image, although a significant improvement could be detected. Patient was given an additional medicine of Moxifloxacin 400 mg once a day and the ATT was continued.

The patient came for a follow up after 3 months of additional therapy. Otoscopy examination gave an image of intact and normal tympanic membrane. Mastoid CT scan showed no fluid the mastoid cavity, and thorax CT scan showed a significant

improvement. Right ear audiometry was rechecked with hearing threshold of 40-50 decibel.

Patient came for follow up in 6 months and one year later without any complaints. Otoscopy examination of right ear, showed a healthy intact tympanic membrane. Right ear audiometry was repeated for evaluation, which had shown a significant improvement although it was not yet normal (Figure 4). No otorrhea in the last one year, although tinnitus was still present.

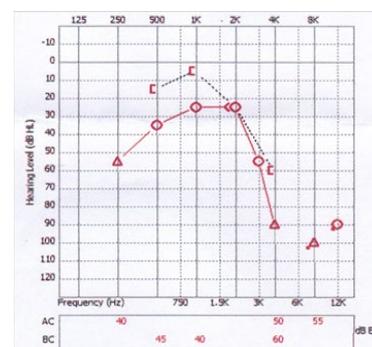


Figure 4. Right ear audiometry post antituberculosis therapy

## CLINICAL QUESTION

How to establish OTM diagnosis?

## REVIEW METHOD

Literature search was carried on in May 2020 using keyword “tuberculous otitis media” through Google Scholar. Selection was based on inclusion criteria as follows: 1) above 18 years old, 2) antitubercular drugs, and 3) managed by surgery. Exclusion criteria was incidence of TOM in children.

## RESULT

The result of of literature search through through Google Scholar, obtained 20 journals which were relevant with the topic.

## DISCUSSION

Tuberculosis is a chronic granulomatous infection caused by *Mycobacterium tuberculosis*.<sup>1</sup> Middle ear infection could occur by way of aspiration through eustachian tube, hematogen spread, through lymphatic vessel from other TB source, or direct infection through tympanic membrane perforation.<sup>1,8,9</sup> Mastoiditis caused by TB complication could happen in repeated middle ear infection through organism penetration via tympanic membrane perforation. Extrapulmonary TB is a predisposing factor of mastoiditis TB through hematogen spread to temporal bone.<sup>1</sup> In this particular case, the most probable route of infection was bacterial aspiration through the eustachian tube, proven by no symptoms of pulmonary TB, and the disease was just diagnosed during pre-operative screening.

There were two development stage of this disease, acute and chronic. In the acute phase, congestion was followed by tympanic membrane thickening, and later the occurrence of tubercles and perforation. While in the chronic phase, it developed slower, accompanied by pain and perforation, where the granulation tissue was pale pink. Necrosis and caseous process had caused bone sequestration and bone destruction.<sup>1</sup>

Up to date, the diagnosis of TOM with mastoiditis remains a significant challenge for otolaryngologists, it required high skill accuracy due to unspecific clinical sign and symptoms.<sup>3</sup> Temporal bones is an organ which is seldom attacked by TB infection. That was one of the reasons of TOM was often misdiagnosed leading to delay in specific treatment.<sup>5</sup> Many a times TOM with mastoiditis had just been discovered several months or years later, especially in patients without any clinical manifestation of the disease. Therefore, high suspicion of TOM is needed, towards patients who live in endemic area, and also towards patients with otorrhea who did not respond to conventional treatment of chronic middle ear infection. The use of

Neomycin and Gentamycin as ear drops could disguise TOM clinical findings, besides both medications have weak antitubercular effect.<sup>1</sup>

There are triad symptoms of TOM, namely otorrhea without pain, multiple tympanic membrane perforations, and facial palsy, although not always all three symptoms are present in each case.<sup>1,6,10</sup> These clinical findings were mentioned for the first time in 1953 by Walmer as TOM signs accompanied by granulation tissue in tympanic cavity and mastoid, and bone necrosis. However, the first TOM case was first described by Jean Louis Petit in 18<sup>th</sup> century.<sup>8,11,12</sup> Hearing disturbance usually appears at the beginning of disease development. Hearing impairment is conductive type as the result of tympanic membrane perforation or the presence of fluid in middle ear cavity. The combination of conductive and sensorineural deafness are rare, it could occur because of labyrinthitis. Facialis nerve palsy was reported in 10-20% cases at the beginning of the disease.<sup>1</sup> It could be concluded that facial nerve palsy without the presence of cholesteatoma could be a warning signal of TOM diagnosis probability.<sup>1</sup> Tinnitus in the elderly is often described as “funny noises”.<sup>13</sup>

The clinical findings in our case, were compliant with what was mentioned in the literatures such as otorrhea without pain, the presence of tympanic membrane perforation, and tinnitus, although there was no facial palsy. The physical examination showed active pulmonary TB without any sign and symptom.

The result of plain skull X-ray was not specific. The skull X-ray showed an opacity or mastoid air cells erosion, similar with other chronic otitis media cases.<sup>1</sup> Temporal bone CT scan was the standard radiological examination for evaluating chronic otitis media. Usually it showed the presence of soft tissue in the middle ear cleft with a normal mastoid air cells, minimal mastoid sclerosis, mucosa thickening of external ear canal, or

soft tissue protrusion into the external ear canal.<sup>1,14</sup> In advanced cases, CT scan might show an extensive damage of temporal bone.<sup>14,15</sup> CT scan was important to find out the degree of temporal bone destruction and to see the complication, such as facial nerve damage, canalis semicircularis erosion or labyrinth ossification.<sup>1</sup>

The gold standard to establish TOM diagnosis are microbiological culture and HPE sample taken from the middle ear and mastoid.<sup>5,6</sup> Positive culture result confirms the presence of bacteria *Mycobacterium tuberculosis*, and confirms the diagnosis of TOM. However, in the ear secret examination, only few cases show positive result. The sensitivity of swab culture for diagnostic was only 20%.<sup>1</sup> Ziehl-Nielsen staining is not a sensitive diagnostic tool, due to minimum count of bacteria in the ear, especially after the use of antibiotic ear drops.<sup>6,10</sup> PCR (*Polymerase Chain Reaction*) method is carried out to detect bacteria *Mycobacterium tuberculosis* in pus or specimen tissue to confirm TB diagnosis.<sup>1,13</sup> Histopathologic examination (HPE) findings of granulation tissue with caseous necrosis, epithelial cell and Langhans giant cells are strong indicators of TB, although false negative could be found in 10% cases.<sup>1,6</sup>

Although TOM was a rare case, TOM could cause serious complications, such as facial palsy, permanent hearing disturbance, retroauricular fistula, labyrinthitis, subperiosteal abscess, tuberculous osteomyelitis, temporomandibular joint disorder, or intracranial complication like meningitis, tuberculomata, otitis hydrocephalus.<sup>2,8,16,17</sup> Severe hearing impairment could occur if the infection invaded the cochlea. Granulation tissue which was spreading into the eustachean tube and pressing the facial nerve could generate facial palsy in 10-20% cases.<sup>1</sup>

Treatment could be commenced with ATT as soon as the clinical and histopathological suspicion aroused to prevent serious

complications.<sup>2,5</sup> ATT duration is minimal 6 to 9 months, varied according to patient's clinical response.<sup>5</sup> Medical resolution for otorrhea take place around 2-3 weeks after commencement of therapy, and the granulation tissue disappears in 1-5 months.<sup>19</sup> Surgery measure is performed for biopsy, subperiosteal abscess drainage, or to remove bone sequestrum. Absolute indication for surgery is the presence of facial nerve palsy, to decompress facial nerve after the failure of conservative therapy.<sup>1,18</sup> A combination of surgery and medications could improve the prognosis.<sup>19</sup> In case of bilateral TOM with bilateral hearing impairment, cochlear implantation is recommended.<sup>1</sup>

There are several surgery procedures according to the patient's condition, such as simple mastoidectomy, radical mastoidectomy, radical mastoidectomy with modification, tympanoplasty, or combined approach tympanoplasty. In a case of safe type chronic suppurative TOM, if conservative treatment failed, simple mastoidectomy surgery could be executed to cleanse pathologic tissue from the mastoid cavity so that the infection could be controlled.<sup>20</sup> While radical mastoidectomy should be performed in dangerous type chronic suppurative TOM, with spreading infection and cholesteatoma. In this operation, mastoid cavity and tympanic cavum are cleansed from all pathological tissue. Modified radical mastoidectomy, also known as canal wall down (CWD) mastoidectomy, is a surgical procedure which involves removal of the posterior (back) wall of the external ear canal to create a common cavity for the mastoid, the middle ear and ear canal. The purpose is to eradicate all pathologic tissues and to prevent intracranial complication, although it could not restore hearing function. Radical mastoidectomy with modification (Bondy operation) is performed on chronic suppurative otitis media with cholesteatoma in attic area, not disrupting tympanic cavum. All mastoid cavity was cleansed and the posterior wall of ear canal was lowered. The purpose of this

surgery was to cleanse pathologic tissue and to retain existing otologic function.<sup>20</sup>

Tympanoplasty is performed on safe type chronic suppurative TOM with more severe damage, or on safe type chronic suppurative TOM which incurable with conservative medicamentous therapy. The purpose of this surgery is to eradicate the disease as well as to repair hearing function. This measure served not only to reconstruct tympanic membrane, but also to reconstruct the ear ossicles.<sup>20</sup> Based on the reconstruction of the ossicles, the reconstruction surgery is known as tympanoplasty type II,III,IV,V. Prior to reconstruction, a thorough exploration of tympanic cavity was executed either with or without mastoidectomy to remove all pathologic tissues.<sup>20</sup> Combined approach tympanoplasty is performed on dangerous type CSOM or on safe type CSOM with extensive granulation tissue. The purpose of this surgery is to eradicate the disease and to repair hearing impairment without radical mastoidectomy.<sup>20</sup>

TOM is a uncommon manifestation of extrapulmonary TB which was rarely reported. High suspicion of TOM is required in patients who did not respond to standard treatment of chronic middle ear infection. The diagnosis is often delayed due to obscure clinical symptoms and the difficulty of biopsy sample taking. The treatment in this case is antitubercular regime, and surgery intervention to cleanse out the secret. Irreversible complication such as permanent hearing impairment could occur in delayed accurate diagnosis.

Our reported case showed how complicated is establishing the diagnosis of TOM, with regards to confusing symptoms and difficulty in obtaining tissue sample. The case is very rare indeed so physician should always be alert of the occurrence of tuberculous otitis media.

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