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Aggressive Tuberculous Otitis Media in a Young Child

ABSTRACT

Objective: The aim of this report is to describe an aggressive case of tuberculous otitis media in a young child and emphasize that surgical intervention and histopathologic studies can be employed to immediately arrive at a definitive diagnosis.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Participants: One

Results: This is a case of a four-year-old boy who had refractory otitis media and erosive CT scan findings, mimicking aggressive CSOM manifestations. Due to unusual intraoperative granulation tissue characteristics, it was initially considered a malignancy, necessitating surgical intervention and biopsy that resulted in a definitive diagnosis of primary middle ear tuberculosis.

Conclusion: This case represents the more severe end of the spectrum of tuberculous otitis media and supports the recommendation that a high index of suspicion, early detection and prompt initiation of treatment are imperative in its management, especially in children with refractory otitis media.

Keywords: *tuberculous otitis media; tuberculosis; middle ear; otitis media; suppurative otitis media*

Tuberculosis (TB) is a rare cause of chronic suppurative otitis media making up only 0.04-0.9% of chronic otitis media cases.¹ Its variable presentation mimics common bacterial middle ear infections such as refractory otorrhea, hearing loss and multiple tympanic membrane perforation.¹⁻⁴ Computerized Tomography (CT) Scan findings include diffuse densities in the middle ear with or without bony destruction of mastoid cavities, ossicles, semicircular canals or the tegmen.^{5,6} Intraoperatively, the typical finding are granulation tissues characterized as pale, whitish to yellowish in color.^{5,6} Due to its variable clinical presentation, TB otitis media is commonly misdiagnosed or undertreated. We present the case of a four-year-old boy with tuberculous otitis media with aggressive manifestations and unusual intraoperative granulation tissue characteristics.

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CASE REPORT

A 4-year-old boy was brought to the ENT outpatient clinic for evaluation of chronic right ear discharge associated with ipsilateral facial nerve paralysis. His six-month history began with watery, non-foul smelling right ear discharge associated with progressive and persistent otalgia. He had been admitted at a tertiary hospital where intravenous antibiotics and chloramphenicol otic drops were administered which allegedly resolved the symptoms. Recurrence of right ear discharge was noted after 3 months, characterized as mucopurulent and associated with otalgia, occasional dizziness, headache, and post-auricular pustule formation. Subsequent decreased hearing decreased activity and sudden onset of right-sided facial weakness prompted consult and subsequent admission. He had no other constitutional symptoms such as fever, cough, night sweats, body malaise or weight loss.

The review of past history claimed complete immunization (based on the Expanded Program of Immunization of the Department of Health), no previous surgeries, no family history of tuberculosis or tumor. He was at par with developmental age, and birth and social history were non-contributory.

Otoscopy revealed bilateral single, central, subtotal (80%) tympanic membrane perforations with erythematous middle ear mucosa and purulent discharge and no mass or granulation tissues. Head and neck examination showed a draining pustule in the right post-auricular area (Figure 1A) and ipsilateral facial asymmetry, House Brackmann Grade 4 (Figure 1B) with no cervical lymphadenopathies palpated. There were no other neurological deficits and an essentially normal systemic examination.

A chest radiograph revealed left pericardiac haziness, interpreted as pneumonia. He also had a slightly elevated white blood cell count of $14 \times 10^9/L$ with neutrophilic predominance while other blood parameters were normal. Temporal bone CT scans showed cortical erosions forming a single mastoid cavity on the right with soft-tissue isodensities within. There was ossicular destruction and bony erosion of the tegmen, petrous portion of the temporal bone, sigmoid sinus and right horizontal semi-circular canal, but the cochlea was preserved and brain parenchyma was normal. (Figure 2A) The left ear only showed opacification of mastoid air cells. (Figure 2B) Pure tone audiometry suggested moderately severe conducting hearing loss on the right and mild conductive hearing loss on the left.

The initial impression was chronic suppurative otitis media with extracranial complications of facial nerve paralysis and subperiosteal abscess on the right. He was started on intravenous ceftriaxone and was scheduled for right canal wall down mastoidectomy under general anesthesia.

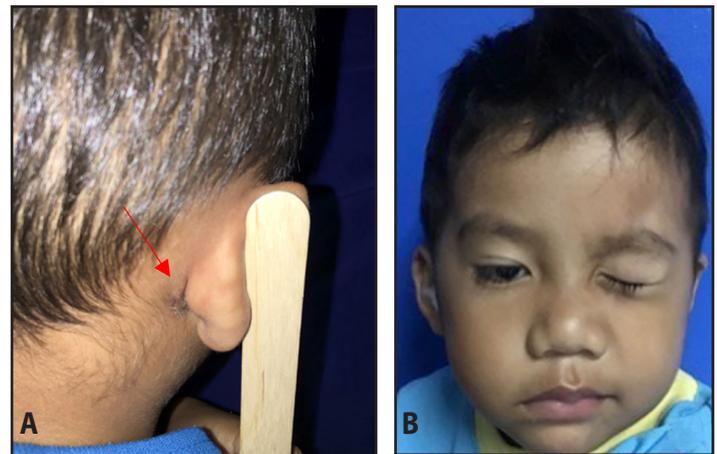


Figure 1. A. Post-auricular subperiosteal abscess (arrow); B. Right facial nerve paralysis, House Brackmann Grade IV. (photos published in full, with permission)

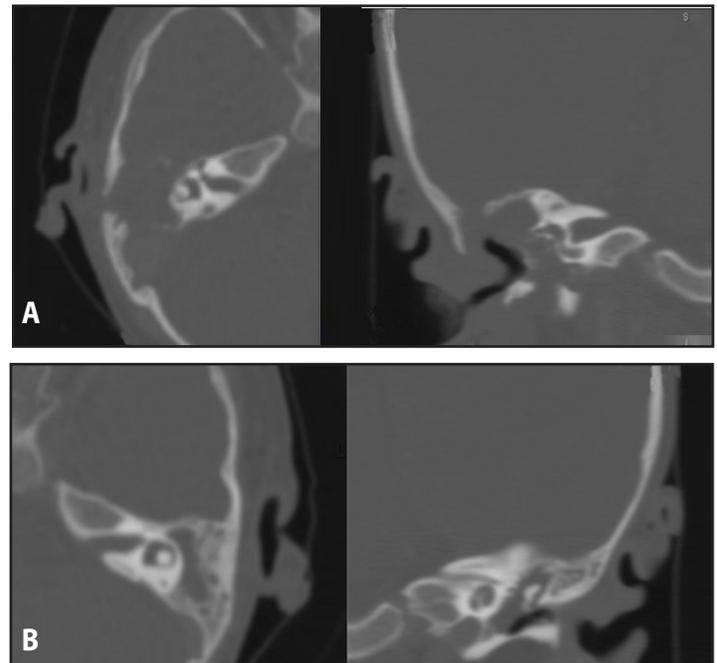


Figure 2. Temporal bone CT scan A. showing erosion of tegmen, sigmoid sinus, lateral semicircular canals and absence of air cells on the right, and B. left mastoid cavity with isodense material within air cells and preservation of the ossicles and semicircular canals.

Intraoperatively, post-auricular subperiosteal dissection revealed irregular cortical breaks with reddish granulation tissues within. (Figure 3A) The spine of Henle was no longer appreciated. Saucerization of the mastoid cavity by drilling further exposed granulation tissue from the tegmen tympani up to the sigmoid area. (Figure 3B) Attempts to dissect and remove the granulation tissues from the tegmen were unsuccessful due to their adherence and the possibility of dural sinus rupture. Considering the possibility of a neoplasm, it was decided

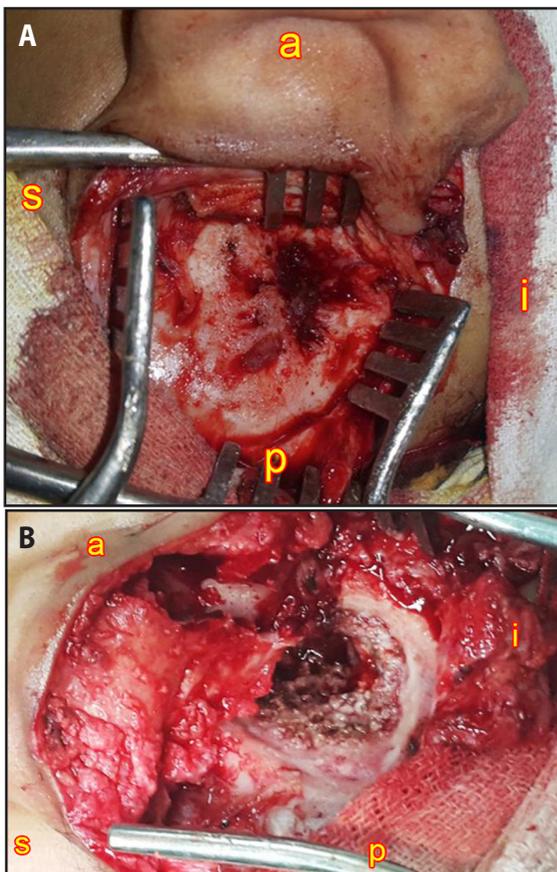


Figure 3. A. intraoperative findings of reddish, beefy granulation tissue upon exposure and B. after attempting excision (a, anterior; p, posterior; s, superior; i, inferior).

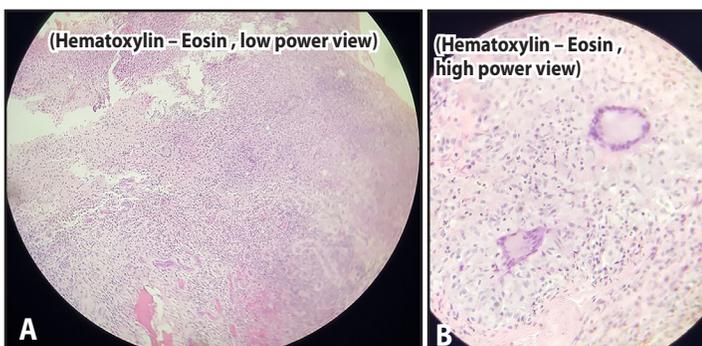


Figure 4. Histopathologic slides of mastoid antrum specimens A. granulomatous inflammation (hematoxylin-eosin, low power view); and B. granulomatous foci with multinucleated giant cells interspersed with lymphoplasmacytic cells (hematoxylin-eosin, high power view).

to obtain samples for histopathology, bacterial and acid-fast bacilli staining and culture and perform a modified meatoplasty and closure.

Final histopathologic results revealed chronic granulomatous inflammation highly suggestive of tuberculosis, described as granulomatous foci with multinucleated giant cells interspersed with

lymphoplasmacytic cells, without evidence of malignancy. (Figure 4) There were no microorganisms and acid fast bacilli seen from the tissue samples on staining and routine bacterial culture results yielded no growth (Lowenstein-Jensen medium was not available in our hospital). The patient was started on dose-adjusted anti-tuberculosis therapy (isoniazid, rifampicin, pyrazinamide and ethambutol) and after 2 weeks of treatment, the right ear discharge resolved. On follow-up 11 months after surgery and upon completion of 6-months anti-tuberculosis medications, both ears were dry and the facial paralysis had improved from House Brackmann Grade 4 to Grade 3 with full eye closure but still with right lip weakness. Unfortunately, they did not follow up again and post-treatment CT scans were not obtained.

DISCUSSION

Tuberculosis is still one of the major health problems in the world especially in low and middle income countries such as the Philippines. Being endemic in the country, there have been numerous documentations of its varied patterns of presentation and extensive spread to extrapulmonary sites. The most common manifestation of extrapulmonary tuberculosis (EPTB) is in the head and neck region, primarily in the cervical lymph nodes with only 2% involving the middle ear.^{4,7}

According to Sens *et al.*, primary TB otitis media affects children more often than adults. It has been classically described as painless otorrhea, multiple tympanic membrane perforations and hearing loss disproportionate to otoscopic appearance.^{1,3,7-9} However, the clinical presentations have changed over the years and have become polymorphic and insidious. Recent reports showed that otalgia is significantly present in most TB otitis media patients, such as the case presented here, and may be due to the pressure caused by granulation tissue occupying the mastoid cavity.^{3,6,8} Tympanic membrane perforations may be multiple in the early stage and coalesce into total perforations at a later stage.⁶ Hearing loss worsens when there is progressive erosion of the ossicles and its bony structures.⁸ According to Sens *et al.*, complications of TB otitis media include facial paralysis, retroauricular fistula, labyrinthitis, meningitis, tuberculous osteomyelitis, intracranial or extracranial abscesses, acute mastoiditis and cellulitis. Facial nerve paresis or paralysis occurs in up to 40% of cases and more frequently in children.^{5,8,10,12}

Tuberculous otitis media mimics chronic suppurative otitis media (CSOM) even on CT scan presentation – soft tissue densities occupying the entire mastoid and middle ear cavities with blunting of scutum.¹ In some studies, there is preservation of the mastoid air cell architecture¹ while others show extensive bony destruction and sclerosis of the



mastoid.² In our patient, the CT scans showed soft tissue attenuation within the mastoid cavity and sclerosis of mastoid air cells. There was ossicular destruction and dehiscence of the lateral semicircular canal and tegmen and blunting of the scutum. These changes are also usually seen in CSOM but may represent late stages of tuberculous infection.

The role of surgery in managing tuberculous otitis media is debatable. It is not essential to treatment and is only reserved for facial nerve decompression, removal of sequestra or improvement of drainage.^{3,8,12} However, it is important to note that surgical intervention facilitates harvesting specimens that pave the way for the definitive diagnosis of TB otitis media. Intraoperatively, the most common finding is a pale, whitish to yellowish soft granulation tissue descriptive of caseous necrosis.^{1,5,6,11} However, there have been reports of granulation tissues adherent to mucosa with abundant dilated blood vessels that eventually led to hematoma formation that might explain the appearance of the adherent reddish soft tissue seen in our patient.^{3,5} The degree of bony erosion and necrosis is variable. Some cases still had identifiable ossicles engulfed in granulation tissue while others had a fistula in the semicircular canals, dehiscence over the facial nerve or erosion up to the mastoid cortex.^{1,3,5,10}

Our case differs from the usual presentation in that the granulation tissue was reddish and closely adherent to the dura so much so that it could not be separated. Together with the aggressive manifestations and extent of involvement, this made us consider the possibility of a soft tissue tumor or neoplastic process. Histopathologic confirmation was the only way to arrive at a diagnosis.

In concurrence with most reported cases of TB otitis media, histopathologic interpretation of the granulation tissue is the most reliable method, showing the presence of *M. Tuberculosis* or epithelioid cells and multinucleated giant cells (Langhans Cells).^{4,7,8} Some recommend polymerase chain reaction (PCR), bacteriologic exam and culture of the discharge, Mantoux and other serologic tests to determine the immune status of the patient. Unfortunately, most of these tests are non-specific and have low yield due to superimposed bacteriologic infection.^{2,5,10,12}

The treatment of TB otitis media is primarily medical. Anti-tuberculosis medications are still the most effective management even for middle ear infection. Most cases respond well, with resolution of middle ear granulations and sometimes full recovery of facial nerve paralysis. The indicator of success or failure is the time interval between onset of the infectious process and initiation of treatment.^{1,8,10,12}

In conclusion, tuberculous otitis media remains one of the elusive causes of chronic suppurative otitis media. Its diagnosis is challenging and its manifestations are evolving into a spectrum ranging from

simple otitis media to tumor-like presentation. Our patient represents the more severe end of that spectrum, who nonetheless responded to treatment well. This case supports previous recommendations that a high index of suspicion, early detection and prompt initiation of treatment are imperative steps in its management especially in children with refractory otitis media.

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