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Case Report

Cutaneous tuberculosis of the pinna: A rare entity

Mohammad Zahir Mohd Sabri[1], *Aynesha Letchmikanthan*[2], *Rosdan Salim*[1]

[1] *Universiti Sains Malaysia, Malaysia*
[2] *Hospital Miri, Malaysia*

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Abstract

Introduction: Cutaneous tuberculosis (CTB) is a rare form of extrapulmonary tuberculosis (TB), accounting for only 1% to 2% of all extrapulmonary cases. It typically affects the face, and to a lesser extent, the neck or chest, with a higher incidence among young women. In Malaysia, TB is endemic; however, CTB involving the pinna is exceptionally rare. Consequently, there is often a delay in diagnosis and treatment due to frequent.

Aim: This case report aims to emphasize the presentation, diagnosis, and subsequent management of cutaneous pinna tuberculosis, supplemented by a review of the existing literature on this disease.

Case study: We present an 83-year-old patient with painful left auricular swelling and deformity and pus discharge for 4 months. His condition did not improve despite prolonged antibiotics treatment and multiple incision and drainage done.

Results and discussion: Examination under anaesthesia, wound exploration extended incision and drainage was performed, revealing auricular cartilage necrosis, tissue biopsy confirmed the presence of *Mycobacterium tuberculosis*.

Conclusions: (1) A non-healing auricular wound despite adequate antibiotics treatment should raise suspicion of TB. (2) Bactec is the most rapid and sensitive culture for tuberculosis. (3) Complications of pinna TB include pain, non-healing wound and permanent ear deformity.

Corresponding author: Rosdan Salim, Universiti Sains Malaysia Health Campus, Jln Raja Perempuan Zainab II, 16159, Kubang Kerian, Malaysia.
E-mail address: rosdansalim@gmail.com

1. INTRODUCTION

Cutaneous tuberculosis (CTB) represents approximately 1.5% of extrapulmonary tuberculosis (TB) cases.[1] Among these cases, involvement of the pinna is exceptionally rare, accounting for only 2.26%.[2] CTB can be classified into true CTB and tuberculids.[3]

2. AIM

We are highlighting the challenges encountered in diagnosing this patient and the subsequent management. The aim is to raise awareness of a rare presentation of pinna infection in the context of CTB. Herein, we present a rare case of cutaneous pinna tuberculosis.

3. CASE STUDY

Our patient is an 83-year-old male who presented with a left auricular swelling persisting for the last 4 months, which has gradually increased in size. He reports experiencing pain and occasional discharge of pus from the surface of the swelling. Despite that, he has not noted any changes in his hearing, tinnitus, or vertigo. He has no known family history of TB but is uncertain about his history of TB vaccination. He denies experiencing any constitutional symptoms, history of exposure to individuals with TB, involvement in high-risk behaviours, or conditions that compromise his immune system.

Upon clinical examination, the patient exhibits a cauliflower-like deformity and swelling of the external ear, primarily affecting the superior aspect of the left auricle, including the anterior and posterior crura, as well as the triangular fossa. The swelling measures approximately 3 × 3 cm and is tender and fluctuant upon palpation. Additionally, there is erythema of the surrounding skin (Figure 1).

Otoscopic examination indicated normal findings in the external auditory canal and tympanic membrane. There were no abnormalities noted in the contralateral ear, and palpation did not reveal any cervical lymphadenopathy.

He had previously completed two courses of oral Augmentin 625 mg TDS before his initial visit to our center, but his condition remained unresolved. Upon evaluation, a tentative diagnosis of left pinna abscess with concurrent perichondritis was established. We proceeded with an incision and drainage procedure on the swelling of the left auricle under local anesthesia. During this procedure, 3 mL of thick, whitish pus was drained, and a Zimmer splint was applied. Shortly after drainage, the swelling visibly reduced in size and the associated pain diminished. Additionally, he received a course of oral ciprofloxacin. Microbiological culture and sensitivity testing of the pus revealed mixed bacterial growth, but no acid-fast bacilli (AFB) were detected.

He returned for a follow-up appointment 1 week later at the outpatient clinic, reporting persistent pain and recurrence of swelling to its original size. A second incision and drainage of the left pinna abscess were performed in the outpatient setting, although no discharge or pus was observed upon incision. Subsequently, he was discharged and completed a total of 6 weeks of oral ciprofloxacin 500 mg twice a day.

Figure 1. Patient: (A) Initial presentation of generalized swelling, erythema, and severe deformity of left pinna; (B) Pus discharge arising from the swelling of the left pinna.

4. RESULTS

Despite completing the antibiotic course, the swelling in the left pinna remained persistent and did not regress. Consequently, he underwent examination under anesthesia for left ear exploration and another incision and drainage procedure in the operating theater. An extended incision was made, encompassing the postauricular region, and another incision was made at the helix (Figure 2). Intraoperative findings revealed a cauliflower-like appearance of the left pinna, minimal pus discharge at the triangular fossa, and thickened, fibrosed overlying skin. Necrosis of the cartilage in the helix and antihelix was observed and subsequently debrided. Otoendoscopy showed thickening of the anterior

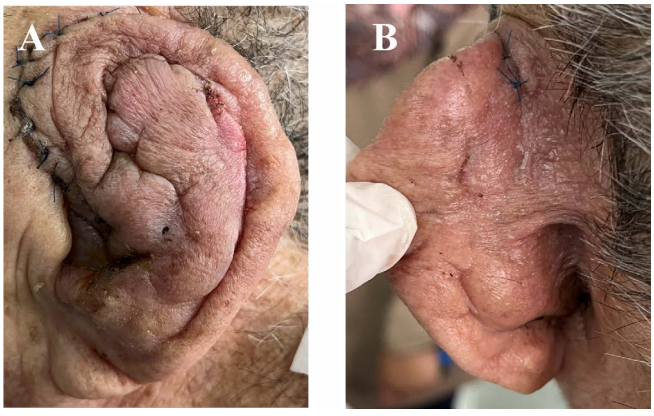

Figure 2. Left pinna 1 week post operation: (A) Reducing swelling of left pinna with mild erythema. Sutures are intact and absence of pus discharge; (B) Surgical excision was done extending to post auricular region showed no signs of recurrent infection.

Figure 3. Postcompletion of anti-TB for 9 months showed resolution of pinna swelling, but permanent disfiguration of the left pinna: (A) lateral view; (B) posterior view.

external ear canal wall with no debris, and the tympanic membrane was intact.

Tissue biopsy using Bactec confirmed the presence of *Mycobacterium tuberculosis*. Subsequently, he was initiated on a regimen of anti-TB medications. The treatment plan included oral Isoniazid, Rifampicin, Pyrazinamide, and Ethambutol (Akurit-4 Tablets, Lupin Ltd, India) for 2 months, followed by an additional 4 months of Isoniazid and Rifampicin. Upon follow-up after completing the anti-TB treatment, the abscess had resolved completely, and the surgical wound had healed well. However, the left pinna still exhibited deformity, resulting in a loss of its normal contour (Figure 3).

5. DISCUSSION

TB is a significant global health challenge and remains a major concern in developing countries. In Malaysia, TB is endemic, with an incidence of 97 cases per 100,000 people as reported in the World Health Organization's Global TB Report for 2021.[4] Among the states in Malaysia, Sarawak has the third-highest number of reported TB cases at 10.6%.[5]

The infectious agent responsible for tuberculosis is *M. tuberculosis*. *M. tuberculosis* is a rod-shaped, non-motile, and unencapsulated bacterium. It belongs to a group of bacteria known as acid-fast bacilli due to its ability to retain carbol fuchsin stain despite exposure to acid decolorization during staining procedures.[6] This characteristic allows *M. tuberculosis* to be easily visualized under a microscope using methods such as the Ziehl-Neelsen stain.[7]

Certain high-risk groups are particularly susceptible to TB infections. These include individuals with recent exposure to TB, smokers, immunocompromised persons (such as those with HIV infection), and those with systemic conditions like diabetes mellitus, chronic renal failure, or malignancies. Injection drug users and individuals living in high-risk, crowded environments such as prisons, nursing homes, or homeless shelters are also at increased risk.[8] HIV-infected individuals are notably vulnerable, with a strong correlation between the HIV epidemic and increased susceptibility to TB reactivation and transmission.[9]

TB can manifest in various parts of the body. Pulmonary TB is the most common form, accounting for over 80% of TB cases, while the remaining 20% are classified as extrapulmonary TB.[6] Extrapulmonary TB refers to TB affecting sites other than the lungs. These can include lymph nodes, pleura, bones and osteoarticular, genitourinary system, gastrointestinal tract, central nervous system, and cutaneous.[10]

CTB can be categorized into true cutaneous TB, where *M. tuberculosis* is identified, and tuberculids, which involve hypersensitivity reactions to bacterial antigens. It further divides into exogenous TB (tuberculous chancre) and endogenous TB (scrofuloderma, lupus vulgaris).[3]

Lupus vulgaris is the most common form of CTB, typically affecting areas such as the head and neck, extremities, and buttocks, often remaining undetected for long periods.[1] Auricular TB is particularly rare, with few reported cases.[9] According to a review by Ramesh et al., only 2.26% of patients with C TB have involvement of the pinna, most commonly affecting the helix or ear lobule.[2] CTB can be acquired through hematogenous or lymphatic dissemination from the lungs or by direct inoculation.[6]

The manifestations of CTB on the pinna can vary, presenting as plaques, hypertrophic lesions, ulcers, or vegetating lesions.[2] In our patient's case, there was initially a plaque-like erythematous lesion, which eventually led to prolonged infection with pus discharge and resultant deformity. Importantly, the infection was confined to the auricle, sparing the external ear canal. Tubercular otitis externa typically develops from nasopharyngeal infection extending through the eustachian tube to the middle ear and eventually involving the external ear canal.[11]

Diagnosing auricular TB involves a comprehensive approach including the patient's medical history, physical examination, presence of active TB elsewhere in the body, histological findings, and response to anti-TB medication.[12]

When *M. tuberculosis* infects the body, the host's immune response begins with macrophages engulfing the bacteria. This triggers the production of cytokines such as interleukins (ILs) IL-2, IL-12, and IL-18, which stimulate interferon-γ (IFN-γ). IFN-γ then activates macrophages to produce tumor necrosis factor (TNF), leading to destruction of the *M. tuberculosis*. Defects in the IFN-γ and IL-12 pathways are crucial factors in susceptibility to *M. tuberculosis* infection.[1] If the mycobacteria persist, granulomas form to contain them, involving the development of giant and epithelioid cells through IL release by macrophages[6]

The differential diagnosis for auricular TB includes subperichondral abscess of the pinna, which typically affects males and presents with recurrent symptoms such as auricular swelling, pain, and pus discharge. Unlike TB, subperichondral abscesses are more common in immunocompromised patients and often associated with trauma or piercing, which this patient did not have.

Other than that, pinna perichondritis is initially suspected. Recurrent partially treated pinna infection leads to

unresolving disease that ultimately develops into cauliflower ear appearance, sparing the ear lobule. Regardless, this condition develops acutely and does not occur over months and is easily treated with oral antibiotics.

Lastly, malignancy of the external ear such as squamous cell carcinoma (SCC) of the pinna is considered as a differential diagnosis. This disease is common in patients aged more than 60 years old, as presented with our patient. The clinical findings of cartilaginous destruction commonly affecting the helix are demonstrated in this condition. In addition, pus discharge may represent necrosis of overlying skin. Despite that, SCC is an aggressive malignancy that spreads to the external ear canal, parotid, mastoid or evidence of facial nerve paralysis or metastases to the cervical lymph nodes.

Diagnosis of extrapulmonary TB can be certain by a battery of tests. The presence of acid-fast bacilli in a culture is the most ideal in demonstrating a diagnosis of CTB. Ziehl-Neelsen staining is a technique used to identify acid-fast bacilli through microscopic examination. This method involves staining mycobacteria with Ziehl-Neelsen dye, which turns acid-fast bacilli a red-purple color. However, the effectiveness of this test depends on the amount of bacteria present, as it may not detect low bacterial loads.[13]

At present, the most rapid and sensitive culture method of identifying mycobacterial infection is Bactec as it can yield a positive culture within a week.[14,15] The Bactec system, which uses the *Mycobacteria* growth indicator tube (MGIT), utilizes liquid media for detecting and culturing *M. tuberculosis*. The liquid medium contains fluorescent indicators and nutrients. As the bacteria grow, they consume the nutrients and produce fluorescence, which is then detected by the system. This allows the Bactec system to identify *M. tuberculosis* even at low bacterial concentrations, making it more sensitive than acid-fast bacilli. Even so, Bactec is expensive, other than its lack of capability to differentiate between a tuberculous and non-tuberculous mycobacterium species in a positive culture.[9]

Although it is not commonly done, we chose to send the intraoperative tissue sample for mycobacterium culture and sensitivity in light of the persistent, long-lasting illness, maintaining our suspicion of TB infection even though the previous acid fast bacilli tests came back as negative.

Concurrent evaluation for pulmonary TB is crucial, as mycobacterial cultures and molecular tests, while specific, can be less sensitive. Tests like Xpert MTB/RIF and Xpert Ultra provide quicker results compared to traditional culture methods for *M. tuberculosis*. TB polymerase chain reaction (PCR) is also utilized as an alternative diagnostic tool.[2] ELISA for anti-tubercular IgM and PCR for *M. tuberculosis* identification can further aid in confirming the diagnosis.[16]

The optimal duration of treatment for extrapulmonary TB, including CTB, is not precisely defined due to limited evidence. However, a multidrug regimen remains the cornerstone of treatment. This regimen typically includes Isoniazid (INH), Rifampicin (RIF), Ethambutol (EMB), Pyrazinamide (PZA), and Streptomycin (STR) as first-line medications. Phase one treatment spans two months with INH, RIF, EMB, and PZA, followed by phase two treatment for four months with INH and RIF.[17,18]

Our patient adhered to the prescribed medication regimen and did not experience any adverse effects. His symptoms resolved completely following completion of the treatment, and there was no recurrence of the disease.

6. CONCLUSIONS

(1) It is crucial to emphasize the diagnosis of CTB of the pinna, especially in countries with a high endemic rate of TB infection.
(2) Features such as a non-healing auricular infection with loss of normal contour despite treatment with antibiotics should raise suspicion for cutaneous TB infection.
(3) A thorough TB work-up, including appropriate diagnostic tests such as mycobacterial cultures, molecular tests like Xpert MTB/RIF or PCR, and serological tests like ELISA for anti-tubercular IgM, is essential for early diagnosis and prompt initiation of treatment for this condition.
(4) Early recognition and management are critical to prevent complications and ensure successful outcomes in patients with CTB of the pinna.

Conflict of interest

All authors certify that they have no affiliations with or involvement in any organization in the subject matter or materials discussed in this manuscript. There is no conflict of interest to declare by all authors.

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Ethics

This case report have been performed in accordance with the Declaration of Helsinki. This case report was reviewed and granted exemption from requiring ethics approval from the ethics committee of Hospital Universiti Sains Malaysia Hospital Jawatankuasa Etika Penyelidikan Manusia. Further information and documentation to support this will be made available to the editor upon request.
Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient. The consent form will be made available to the editor if requested and will be treated confidentially.

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