CASE REPORT

Atypical presentation of aural tuberculosis with complication

Anup Singh,[1] David Victor Kumar Irugu,[1] Hitesh Verma,[2] Alok Thakar[2]

[1]Department of ENT, All India Institute of Medical Sciences, New Delhi, India
[2]Department of ENT and Head and Neck Surgery, All India Institute of Medical Sciences, New Delhi, India

Correspondence to
Dr David Victor Kumar Irugu, drdvki2776@gmail.com

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SUMMARY

Tuberculosis involving mastoid and ear is an uncommon entity presenting with myriads of non-specific features and difficult to diagnose, being a paucibacillary condition. The involvement of otomastoid compartment is hypothesised to be of haematogenous origin. Rarely it can spread directly via tympanic membrane perforation or via reflux through eustachian tube. The usual picture of presentation tends to be one of indolent ear infection not responsive to usual antibiotic treatment and symptomatology being out of proportion to examination findings. We present a case of aural tuberculosis presenting with zygomatic and Bezold abscess without other symptoms, and the usefulness of GeneXpert test in mycobacterial detection in such paucibacillary conditions.

BACKGROUND

This report fulfils the following two requirements as mentioned in the list of the cases the journal is interested in publishing:

- Unusual association of diseases/symptoms.
- Unusual presentation of more common disease/injury.

CASE PRESENTATION

A 38-year-old man presented to our outpatient department with a history of left side postaural swelling of a 5-month duration. The patient denied any other significant ear/nose/throat symptomatology. The patient had some vague aural fullness and some sensation of wetness in left ear. On examination, the patient had a fluctuant swelling involving left mastoid tip and adjacent suboccipital area (figure 1).

Otoscopy revealed dull tympanic membrane on left side with polypoidal tissue filling up posterosuperior quadrant and pure tone audiometry showed mild conductive hearing loss on left side.

INVESTIGATIONS

High resolution computed tomography (HRCT) temporal bone 0.6 mm axial and coronal cuts showed soft tissue density filling up left middle ear and mastoid cell system, with intact septations, ossicles, semicircular canals and facial canal (figure 2). Mantoux test, blood profile, erythrocyte sedimentation rate, viral markers and chest X-ray were normal.

DIFFERENTIAL DIAGNOSIS

1. Suppurative lymphadenitis (possibly tubercular).
2. Otogenic deep neck space abscess (Bezold abscess).
3. Infected branchial cyst.

TREATMENT

The patient was taken up for mastoid exploration under general anaesthesia. On table assessment confirmed the preoperative examination under microscope (EUM) findings. Via a postaural incision, the abscess was accessed and around 15 mL thick pus was drained. Intraoperatively, the entire mastoid and middle ear were found to be full of pale granulations along with liquefied caseous tissue (figure 3). Pressing over the neck abscess area caused purulent material to pool inside mastoid tip area demonstrating direct communication of mastoid and neck pathology, which was traced and the area of communication was found medial to digastric ridge. Drilling of zygomatic root area revealed localised pus collection involving zygomatic cells, which was drained. A canal wall down mastoidectomy was done to remove the entire disease and the granulation tissue and pus discharge were sent for Gram stain, acid-fast bacilli staining, culture sensitivity and GeneXpert along with histopathology.

OUTCOME AND FOLLOW-UP

Postoperative period was uneventful. Postoperative reports revealed necrotising granulomas and GeneXpert positive (Rifampicin (RIF) sensitive) for tuberculosis. Postoperatively, pending culture report, the patient was kept on amoxicillin–clavulanate combination for 3 weeks but ear discharge continued. The culture report (Bactec) after 3 weeks revealed no growth, but in view of non-improving patient condition with strong clinical and supporting ancillary test results, patient was put on antitubercular therapy. Within 2 weeks of starting therapy, patient experienced significant subjective improvement in ear and general condition. At 1 month, the granulations in cavity were reduced to minimal and at 2 months, the cavity was well epithelised without any residual granulations or discharge (figure 4).

The patient completed a 9-month completion regimen with rifampicin and isoniazid after receiving 3 months of intensive phase with rifampicin, isoniazid, pyrazinamide and ethambutol, and remains disease free at 1-year follow-up post therapy completion.

DISCUSSION

Tuberculosis involving otomastoid compartment is a rare condition. The presentation with variety of

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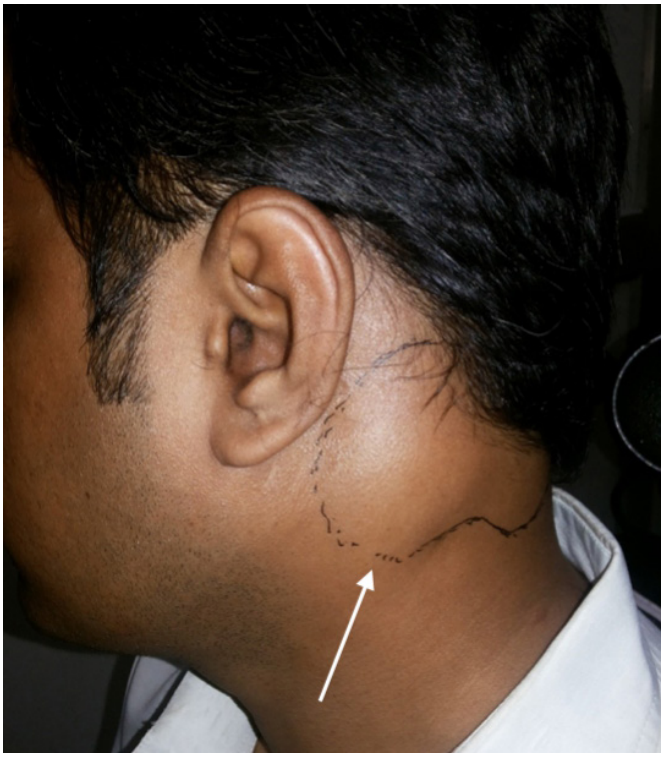

Figure 1 Arrow showing the Bezold abscess outlined on left side.

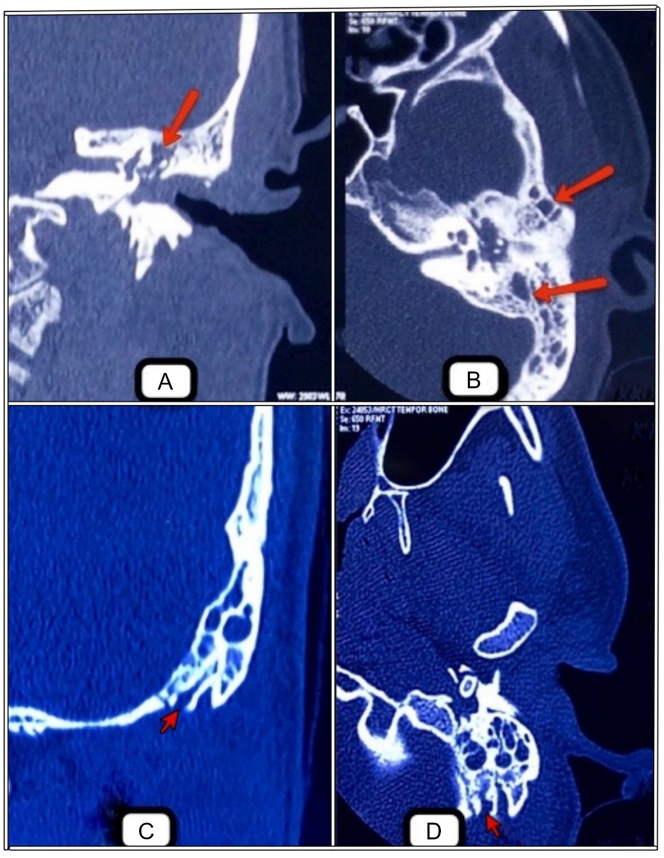

Figure 2 High-resolution CT temporal bone (A and B) showing soft tissue (red arrows) involving middle ear and mastoid with intact mastoid cell system, and (C and D), showing cortical erosion (small red arrows) of posteromedial part of mastoid tip, the putative site of egress of infection leading to abscess formation in the soft tissue of the neck.

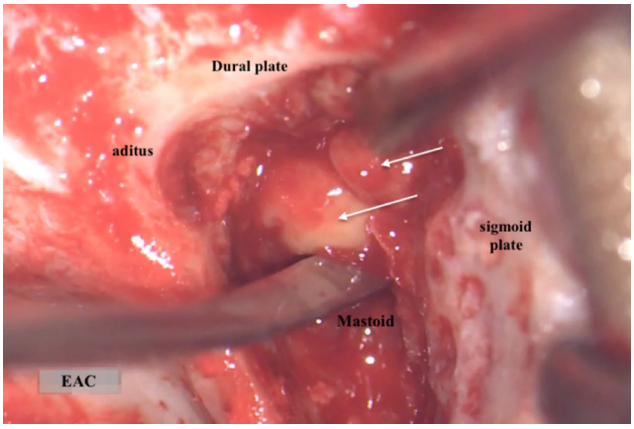

Figure 3 Intraoperative picture showing pale granulations (white arrows) filling up the mastoid. EAC, = external auditory canal.

non-specific signs and symptoms makes it a diagnostic dilemma. To the best of our knowledge, this is the first reported case of aural tuberculosis presenting with a Bezold and zygomatic abscess without any other significant symptomatology.

Bezold abscess refers to collection along the posterior belly of digastric muscle formed as a result of pus trickling along the fascial plane from eroded medial aspect of mastoid tip in a case of suppurative mastoiditis, the apparent extension to skin surface being prevented by the resistance offered by sternocleidomastoid, trapezius and splenius muscles.[1] Zygomatic abscess as a complication of otitis media, first described by Bezold in 1908 as an exceedingly rare occurrence, usually occurs in well pneumatised zygomatic root with ample of zygomatic root cells, which get involved in continuity with the rest of the mastoid cell system.[1]

The most common route of spread of infection to ear remains haematogenous from focus of infection elsewhere in body. Significant changes in epidemiology and presentation of tubercular otomastoiditis have been observed post BCG vaccination and antitubercular chemotherapy era.[2] The classic picture of aural tuberculosis is described as a painless chronically draining ear,

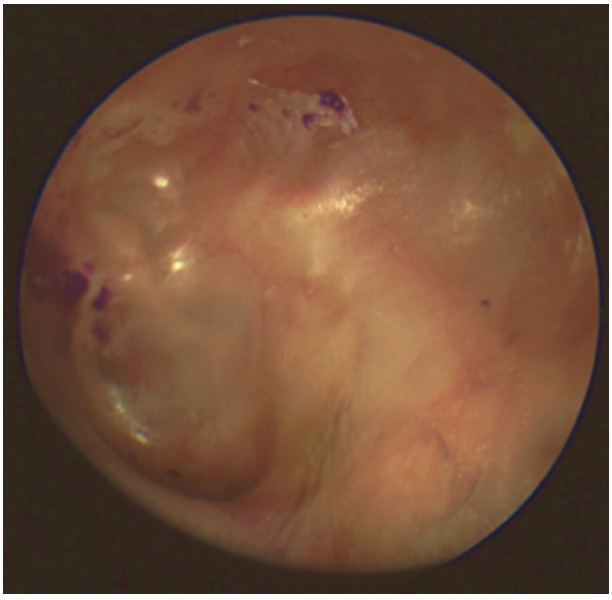

Figure 4 Postoperative mastoid cavity after 2 months of antitubercular therapy.

not responsive to topical and systemic antibacterials, with nature of discharge initially being serous and later on becomes mucopurulent. Hearing loss is usually out of proportion to the patient symptomatology and may be conductive (90%), sensorineural (~8%) or mixed (~2%).[3] The entire tympanomastoid compartment is involved by profuse pale granulations, which appear spongy and sprout up under tension on opening up of mastoid cells during surgery, considered to be a fairly characteristic intraoperative finding.[3 4] Left ignored, the tubercular process may lead to the involvement of underlying bone of mastoid compartment with osteomyelitic changes and irreversible changes in bony labyrinth. Isolated promontorial/cochlear fistulae are more common with tubercular otitis than with non-tubercular infectious aetiology.[5]

Discordance between symptomatology and clinical findings remains a constant relationship (as evident in our case also) characterising tubercular otitis media and incidence of complications like facial palsy (16% in adults and 35% in children) and sudden or progressive sensorineural hearing loss are especially noticeable with tubercular otitis media (TOM),[6] and can be presenting features without otorrhoea as well.

Aural tuberculosis is a paucibacillary disease. More often than not, the clinician has to rely on supportive evidences to start treatment, given the difficulty to demonstrate tubercular bacillus in the small amount of tissue obtained for processing. AFB positivity of stained tissue remains low (2%–14%).[2 6] Histopathology shows necrotising granulomas more often, and culture takes a long time for positivity with a low yield.[7]

GeneXpert mycobacterium tuberculosis (MTB)/RIF test is a PCR-based assay that can detect presence of MTB DNA (*rpoB* gene) with a sensitivity of 131 colony forming units of MTB/mL within 2 hours and can give idea about resistance to rifampicin.[8] It has an important adjunct value in making diagnosis and starting treatment, but further prospective studies are needed to establish the value of GeneXpert as rapid diagnostic tool in a paucibacillary site like otomastoid compartment. It carries a high sensitivity (>99%) in detecting rifampicin resistance, however, for monitoring purposes, the conventional microscopy remains the standard.[9]

Its important to start antituberculosis treatment timely and on the basis of contributory finding rather than waiting for culture report, since the later may take a long time and meanwhile result in disease progression. Involvement of otomastoid compartment should be treated like bone tuberculosis with a total duration of treatment up to 9–12 months. The surgical treatment in today's era should be reserved for complications including subperiosteal abscess, facial palsy and sequestrum removal.[10]

Learning points

- Tubercular otomastoiditis can present with subperiosteal abscess with masked ear symptomatology and needs high index of suspicion to diagnose.
- Paucibacillary nature of disease and small amount of obtainable tissue makes demonstration of organism in tissue difficult.
- Treatment should be initiated on clinical grounds. Ancillary tests and culture can aid confirming diagnosis and guide therapy.

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