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Case Report

Temporal Bone Osteomyelitis Masquerading as Malignancy: A Diagnostic Challenge

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Xanthogranulomatous osteomyelitis is a rare chronic inflammatory disorder. Until now, it has only been reported in long bones. To the best of our knowledge, it has never been reported in temporal bone. We present the case of this rare disease in a 64-year-old male involving the temporal bone, presenting with ear pain, discharge, decreased hearing, and granulation tissue in the external auditory canal, mimicking malignancy clinically and radiologically. The patient was unresponsive to medical management and was taken up for surgical debridement, followed by treatment with systemic and topical antibiotics, with a successful outcome. As this disease has not been reported in the literature yet in the temporal bone and mimics malignancy, it must be differentiated on histopathology to establish a definite diagnosis and provide appropriate management. A long-term follow-up is also necessary to recognize the clinical behavior of this disease, as no treatment protocol has been established yet.

KEYWORDS: Osteomyelitis, temporal bone, otology

INTRODUCTION

Xanthogranulomatous osteomyelitis (XO), a rare chronic inflammatory disorder, is clinically characterized by pain, fever, and leukocytosis. Xanthogranulomatous inflammation has been reported in various organs like the gallbladder, pancreas, fallopian tube, ovary, epididymis, testis, kidney, prostate, and salivary gland[1,2] and rarely in the brain, lung, and bone.[2]

Bony involvement of xanthogranulomatous inflammation is termed as XO. Grossly, it presents as a mass-like lesion that extends to enclose adjacent tissues, mimicking malignancies.[3] Histologically, the disease is characterized by the collection of foamy macrophages along with polymorphonuclear leukocytes, plasma cells, and polyclonal lymphocytes in a mosaic-like pattern.[4,5] Histological differentiation of this pathology from malignancy is important to avoid extensive surgical interventions.[5] Although few cases of XO in long bones have been reported in the literature, to the best of our knowledge, it has never been reported in the temporal bone before. Here, we report the first case of temporal bone XO in a 64-year-old male who presented with ear discharge, pain, and decreased hearing. He did not respond to conservative management with systemic antibiotics and multiple inconclusive biopsies, so the patient was planned for surgical debridement. The histopathological report came out to be XO. Postoperatively, it was managed with systemic and topical antibiotics and regular wound cleaning.

CASE PRESENTATION

A 64-year-old male presented in the otorhinolaryngology outpatient department (ENT OPD) with complaints of left ear discharge, pain, and decreased hearing for the last 6 months. On examination, the external auditory canal (EAC) was completely filled with granulating mass up to the external meatus. The patient had moderate mixed hearing loss on pure-tone audiometry. High-resolution computed tomography of the temporal bone (Figures 1A and B) revealed hypodense soft tissue in the middle ear cavity extending to the attic, aditus, antrum, and hypotympanum, extending laterally into the EAC with erosion of its floor and anterior wall. A biopsy was taken twice from the granulation tissue under local anesthesia, which showed non-specific chronic inflammatory pathology. The patient received antibiotics (piperacillin–tazobactum 4.5 g IV thrice a day [TDS] plus oral ciprofloxacin 750 mg twice

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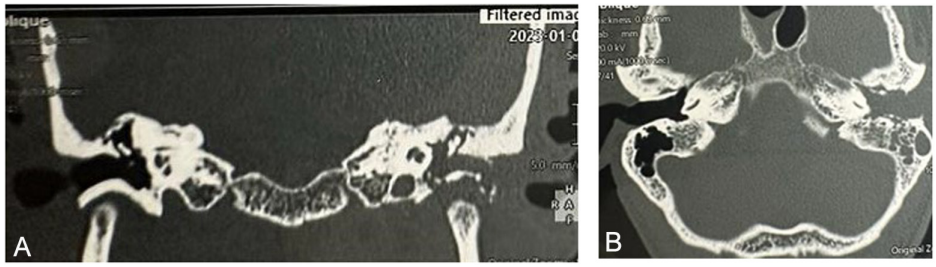

Figure 1. Coronal (A) and axial (B) high-resolution computed tomography temporal bone images showing hypodense soft tissue filling the left middle ear cavity and mastoid air cells, with extension to the external auditory canal eroding its floor and anterior wall.

a day [BD]) for 2 weeks, but there was no clinical improvement in signs and symptoms. In view of no response, the patient was planned for surgical debridement. The patient was then taken up for mastoid exploration via a post-auricular approach under general anesthesia. After soft tissue dissection and cortical mastoidectomy, the middle ear, attic, aditus, antrum, hypotympanum, and EAC were found to be filled with granulation tissue, which was removed and sent for histopathology. The anterior wall of the EAC was eroded, and granulation tissue was reaching up to the glenoid fossa, which was cleared. The facial ridge was lowered up to the level of the facial nerve (Figure 2). No ossicle was present except for the stapes footplate. Middle ear sealing was done with conchal cartilage and temporalis fascia, as the middle ear mucosa was found to be unaffected. Concho-meatoplasty was done, and the wound was closed in 2 layers. The histopathological report showed sheets of foamy macrophages, along with dense chronic inflammatory cell infiltrates comprising neutrophils, plasma cells, lymphocytes, and giant cells compatible with the diagnosis of XO (Figure 3). Necrotic bone was seen with the loss of osteocytes. The patient was kept admitted postoperatively for 48 hours and discharged under stable conditions. He was kept on a regular monthly follow-up with an epithelized mastoid cavity with minimal granulation tissue after 2 months and a completely healed cavity after 3 months (Figures 4 A and B).

The informed consent was obtained from the patient for publication of this case report. This study was approved by Ethics Committee of Government Medical College, Patiala (Approval No: 9(310)2023/14830, Date: May 16, 2023).

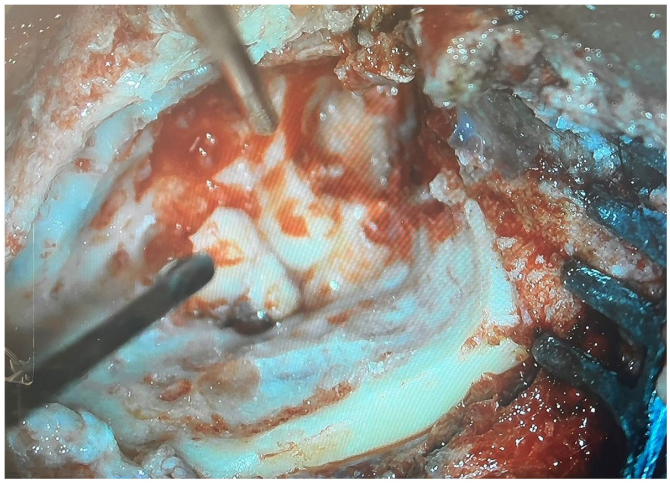

Figure 2. Intraoperative microscopic picture showing clearance of the left mastoid cavity from granulation tissue during modified radical mastoidectomy.

DISCUSSION

The XO of bone was first reported by Cleto Cozzutto in 1984 in 2 cases involving the rib and tibia. Bone involvement is less common in this condition, and predominantly it is observed in soft tissues. It has been described in the gallbladder, kidney, lung, and gastrointestinal and urogenital tracts.[6-8]

In 2019, Solooki et al[9] published a case report and review of literature of 20 cases of XO, all in long bones, along with their own case. The most common bones involved were the tibia (8), femur (5), rib (3),

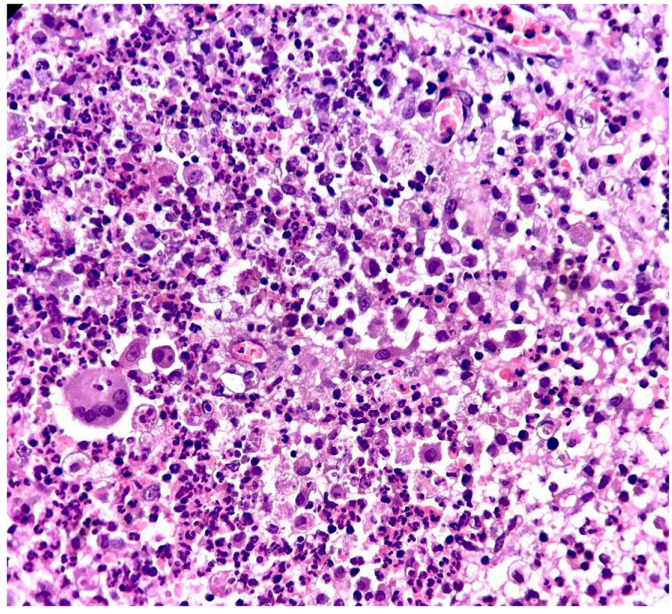

Figure 3. Microphotograph showing dead bone with mixed inflammatory cell infiltrate and foamy histiocytes (H&E, 40×).

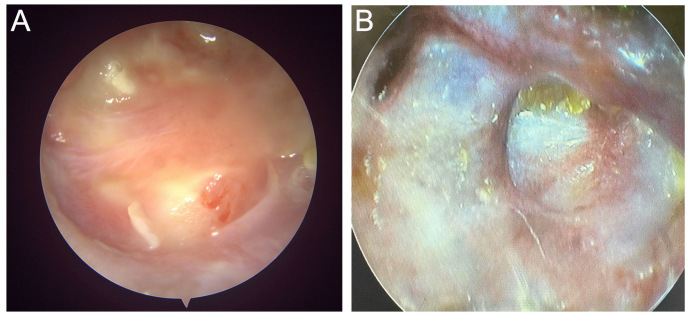

Figure 4. Postoperative endoscopic picture showing a well-healed mastoid cavity with minimal granulation tissue at 8 weeks (A) and complete healing at 12 weeks (B).

humerus (3), and ulna (2). Involvement of metatarsals and metacarpals has also been reported in 1 case each. Temporal bone involvement was not seen. They showed a male predominance (15/21) with no characteristic age distribution. Pain and swelling at the involved site were the most common presenting features, and 3 patients also had a preceding history of trauma. Radiology showed lytic lesions in almost all cases, and 17 out of 21 patients had unifocal lesions, while only 4 had multifocal disease.

The pathogenesis of this disease is attributed to the delayed type IV hypersensitivity reaction of cell-mediated immunity. Even though xanthogranulomatous lesions of several organs, such as the kidneys and the gastrointestinal system, have been said to be associated with bacterial infection, their association with bony involvement has not been established yet.[4] The clinical and radiological presentation of XO mimics malignancy. In our case also, the patient had severe otalgia, otorrhea, hearing loss, and EAC granulations with extensive bony erosions on computed tomography suggestive of malignancy. Thus, a histopathological diagnosis is essential to differentiate it from carcinoma.[4,10-13]

Clinical, radiological, and histopathological findings need to be correlated together to make the diagnosis of XO. However, histopathology is the gold standard in the final diagnosis of the disease.

The differential diagnosis of xanthogranulomatous inflammation includes Langerhans cell histiocytoses, Erdheim–Chester disease, chronic recurrent multifocal osteomyelitis, xanthoma, infiltrative storage disorder, malakoplakia, fibrohistiocytic tumor, and metastatic renal cell carcinoma.[14] In the temporal bone, skull base osteomyelitis and tuberculosis can also be considered as differentials due to the presence of granulation tissue. Relapsing polychondritis can also be considered as one of the differential diagnoses as it relapses and causes progressive destruction of the cartilage tissue.[15] The patient can present with ear pain and hearing loss.

As XO is a rare pathology, no standard treatment protocol exists for the disease. Treatment with curettage and bone grafting has been reported in most cases of long bones. Antibiotics have also been given along with surgical procedures in 2 patients, and in 3 patients, treatment was based completely on broad-spectrum antibiotics with regular follow-up, as reported by Solooki et al.[9] In our case, surgery was done initially, and following the histopathology report, oral cephalosporin (cefuroxime axetil) was prescribed for 4 weeks along with topical antibiotic eardrops. At the last follow-up of 8 weeks, there was more than 90% resolution of granulation tissue and complete resolution of ear pain and discharge.

In conclusion, xanthogranulomatous osteomyelitis is a rare disease seen mostly in long bones, with the first time being reported in the temporal bone. As this disease is unusual in the temporal bone, its awareness and correct diagnosis by histopathology, especially to differentiate from malignancy, are crucial in the appropriate management of this disease. The treatment is debridement, followed by systemic and topical antibiotics. A long-term follow-up is necessary to understand the clinical behavior of this rare disease.

Ethics Committee Approval: This study was approved by Ethics Committee of Government Medical College, Patiala (Approval No: 9(310)2023/14830, Date: May 16, 2023).

Informed Consent: Informed consent was obtained from the patient who agreed to take part in the study.

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