



## Case report

## Primary tubercular osteomyelitis of zygoma: A literature review and case report

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## ABSTRACT

**Purpose:** This study presents a rare case of primary tubercular osteomyelitis of the zygoma, and addresses how combined surgical and medical treatments are effective managing rare presentations of tubercular osteomyelitis in facial bones.

**Methods:** A 57-year-old man presented with pain and purulent discharge from the right malar region following trauma. Initial treatments with empirical antibiotics had poor response, hence further investigations were done. Comprehensive diagnostic workup included lab tests, Computed tomography with contrast, and histopathological examination. The patient underwent surgical debridement of the zygomatic bone through an intraoral approach, and debrided tissue was sent for microbiological and histopathological examination, which confirmed tubercular osteomyelitis. Post-surgery, the patient was treated with a four-drug anti-tubercular regimen.

**Results:** Following anti-tubercular therapy, the patient showed significant improvement, with no signs of disease recurrence after a year of follow-up.

**Conclusion:** Primary tubercular osteomyelitis of the zygoma is extremely rare but should be considered in differential diagnoses of facial swellings and discharging sinuses. This case underscores the importance of a multidisciplinary approach in treating rare presentations of tubercular osteomyelitis.

## Introduction

Tuberculosis (TB) is amongst the most ancient infectious conditions globally in terms of preventable morbidity and mortality rates. The World Health Organization (WHO) emphasized in its 2021 global TB report that each year, approximately 10 million people contract TB, leading to 1.5 million deaths [1]. High-risk groups for tuberculosis include individuals who are immunosuppressed, malnourished, of low socioeconomic status, alcoholics, healthcare workers, and individuals that live in TB-endemic areas.

One in five TB cases manifests as Extrapulmonary tuberculosis (EPTB), in concordance with data from India where 16 % of newly reported cases were of EPTB [2]. A slow-growing aerobic bacillus called *Mycobacterium tuberculosis* is the causative agent and can enter a quiescent state, complicating detection and treatment. Its intricate interaction with the host results in a spectrum of disease states, from latent infection to active clinical disease, which are not fully understood [3]. Transmission occurs through particles that are airborne, released by coughing, talking, or sneezing of an infectious TB patient.

Among various extrapulmonary types, oral and head & neck TB contribute to approximately 10 % to 15 % of the cases [4]. The orofacial manifestations of TB may develop as an array of ulcers, gingivitis, periapical granulomas, or sialadenitis. But the rarest manifestation of orofacial TB is tuberculous osteomyelitis of the jaws, accounting for less than 2 % of all skeletal TB [5], particularly in the mandible. Osteomyelitis is rarely seen in bones of the midface due to ample vasculature. Infection can spread through various routes, including direct transfer from infected sputum, through openings in the oral mucosa related to tooth eruption or extraction sockets, or to underlying bone. Tuberculosis affecting the jaw leads to gradual bone necrosis and the development of a sub-periosteal abscess, known as "lumpy jaw," characterized by a swelling that is soft and painless. Rupture of this abscess may lead to the formation of single or multiple sinuses either within the mouth or externally [6].

Small, flat bones of the face, nasopharynx, nose, maxilla are uncommon sites for TB, but the rarest entity is primary TB of zygoma. Therefore, we are presenting a case study that highlights the unusual occurrence of primary zygomatic tubercular osteomyelitis, along with

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its characteristic features and treatment approach. This case aims to educate clinicians about uncommon presentations of a typically encountered condition.

### Case presentation

A 57-year-old gentleman reported to our facility with the complaints of pain and purulent discharge from the right malar region since previous 15 days. The patient had sustained trauma three months prior; he was hit by an animal. There was no significant family history reported, nor did the patient have any co-morbidities or any prior history of TB, but the patient belonged to a low socio-economic background and resided at a demographic area with concerning TB rates. Following the trauma, the patient was first treated elsewhere, where drainage of hematoma over right malar region was done. A month later the patient had an episode of bleeding from the same region, which was managed by exploring the wound and ligating the bleeding vessel, along with application of bone wax. Two weeks later the patient developed pain and pus discharge from the right malar region, and fifteen days had passed before the patient visited our facility.

At the time of reporting, the local examination revealed a sinus tract having undermined edges present over the inferior aspect of the lateral canthus of the right eye, with an overlying defect of size  $2 \times 1.5$  cm (Fig. 1). There was no clinically palpable lymphadenopathy evident. Intra-oral examination showed a tender swelling over the right malar region without pus discharge or associated tooth mobility.

### Workup

Lab tests showed a haemoglobin level of 11.6 gm%, an erythrocyte sedimentation rate (ESR) of 38 mm/hr, and a total leukocyte count (TLC) of 6100 cells/mm<sup>3</sup>, with 58 % polymorphs and 40 % lymphocytes. Empirical antibiotic coverage did not alleviate the symptoms, nor showed any reduction in pus discharge, thus further investigations were done. A computed tomography (CT) scan of the orbit and paranasal sinuses revealed that the skin and subcutaneous tissue over the right zygomatic region and right infraorbital region showed thickening. Irregular skin ulceration was also noted. Minimal fluid collection and a few non-enhancing foci were noted in the deep subcutaneous tissue. The underlying right zygomatic bone and the zygomatic arch also show erosion. All the above findings, along with cortical irregularity and a few cortical breaks, were suggestive of chronic osteomyelitis of the right zygomatic bone with overlying cellulitis. The pus from the sinus was sent for culture and AFB, the reports did not provide definitive results.

The initial diagnosis was determined to be chronic osteomyelitis of the zygoma, and the patient was planned for debridement and curettage.

### Treatment and follow-up

An intraoral approach was taken from the right maxillary vestibule for exposing the zygoma and zygomatic arch, and debridement of the bones was done until healthy bleeding edges were elicited. A perifistular incision was given near the lateral canthus of right eye, and after

checking the patency of the sinus tract, thorough debridement and curettage was done. Wound margins were refreshed and closure of the skin was done using 4-0 Prolene suture. The debrided bone along with the soft tissue (Fig. 3) was sent for histopathological and microbiological examination, revealing a granulomatous lesion with caseation, indicating the possibility of tuberculosis.

Postoperatively, a chest x-ray was done, which did not show any fibrocavitary lesions or any signs of active pulmonary tubercular infection. CBNAAT test was performed, reporting MTB negative. GeneXpert assay showed sensitivity to first-line anti-tubercular drugs, leading to the initiation of Anti-Tubercular Treatment (ATT) involving a four-drug regimen of Rifampicin (RIF), Isoniazid (INH), Ethambutol (EMB), and Pyrazinamide (PZA). For the initial two months of treatment, an intensive phase of RIF, INH, EMB, and PZA, followed by a seven-month continuation phase of INH and RIF.

The patient responded well to the treatment. Following the completion of ATT therapy, the patient attended routine follow-up appointments. After over a year of treatment, the patient remains asymptomatic, with no signs of disease recurrence.

### Discussion

Bone and joint tuberculosis is a known disease since ancient times, with signs of osteoarticular TB identified in Iron Age remains from Asia, Egyptian mummies, and European skeletons from the Middle Ages through polymerase chain reaction (PCR) analysis, or histology [6]. *Mycobacterium tuberculosis* is predominantly responsible for mycobacterial osteomyelitis and arthritis globally. Due to the AIDS epidemic, there was a rise in rare nontuberculous mycobacterial (NTM) skeletal infections during the 1980s and 1990s. NTM infections have been linked to puncture injuries, orthopedic surgeries, and intravesical bacillus Calmette-Guérin (BCG) treatments [7].

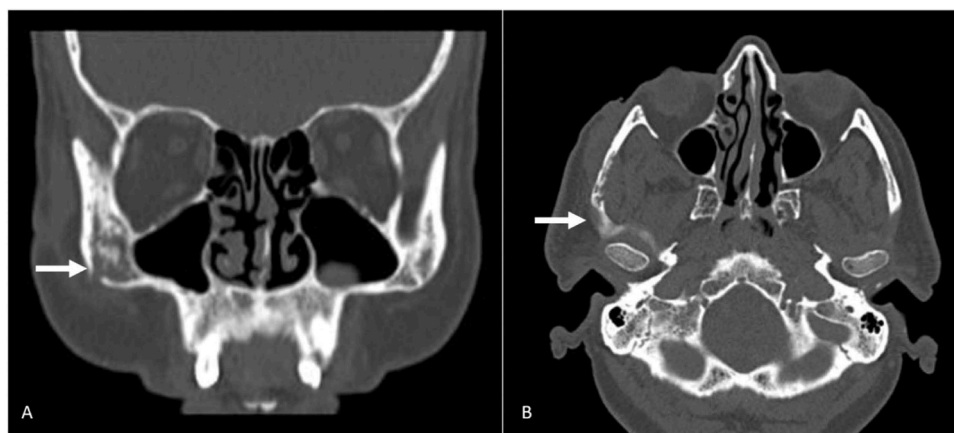
Spinal tuberculosis (Pott's disease) accounts for 50 % of tuberculous osteomyelitis, followed by metaphyses of long bones, but tuberculous osteomyelitis of small flat bones particularly of the face has an exceptionally low incidence rate. In a series of 23 cases of tuberculosis of head and neck analysed by Penfold and Revington, only a single case of tuberculous osteomyelitis of zygoma was encountered [8]. Osteomyelitis and arthritis in the background of tuberculosis typically result from the reactivation of bacilli of the original infection [9], although the patient discussed in the present case did not report any previous history or exposure, nor any clinical or radiological evidence of pulmonary TB.

Typically, the initial symptoms of facial bone tuberculosis manifest as fluctuant swelling with or without a discharging sinus. However, in this particular case, the primary symptom was swelling around the zygomatic bone. Other symptoms may include neuralgia and non-specific headaches. Table 1 depicts cases in literature that have reported primary tubercular osteomyelitis of the zygomatic bone, and their line of management.

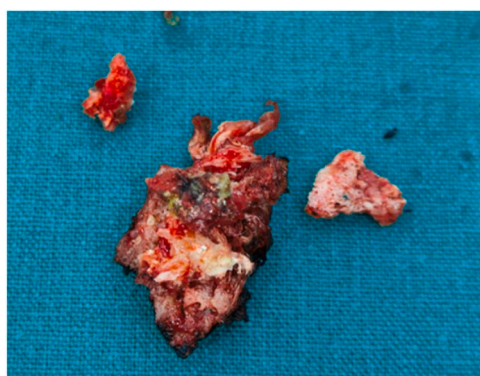
Cytology confirmed the diagnosis of the current case, corroborated by osteolytic lesions observed in radiology. In cases where radiological and histopathological evidence is inconclusive, additional tests such as enzyme-linked immune sorbent assay (ELISA) for detecting IgG and IgM



Fig. 1. Swelling with sinus tract present over inferior aspect of lateral canthus of right eye.



**Fig. 2.** CT scan of the zygomatic arch in (A) coronal section, and (B) axial section showing erosion of bone.



**Fig. 3.** Debrided necrotic bone from right zygoma.

antibodies against specific mycobacterial antigens, as well as GeneXpert/CBNAAT may be employed [15].

Extrapulmonary tuberculosis often requires different treatment regimens compared to pulmonary tuberculosis due to the diverse sites and complexities of infection. While PTB typically follows a standard 6-month treatment regimen, EPTB may necessitate prolonged therapy of 9 to 12 months due to the difficulty in achieving effective drug penetration and ensuring complete eradication of the infection. During the intensive phase, these drugs are often administered daily or five times per week for eight weeks. In the continuation phase, the regimen can shift to thrice-weekly dosing [16]. The mainstay of treatment is conservative, focusing on ATT administration, while surgery is reserved for cases having intracranial involvement, extensive destruction, or secondary infection. The patient continues with regular follow-up and is disease free till date, without any recurrence or discomfort.

**Table I**

Reports of primary tubercular osteomyelitis of the zygomatic bone and their management.

Sr. No.	Author	Year	Patient A/S	Primary/prior history/ history of exposure	Bone involved	Treatment	Follow-up
1.	Sachdev [10]	1993	4/M	Father being treated for pulmonary TB	Right zygomatic bone	Surgical + Medicinal: Saucerization of ulcer under GA	Complete healing of wound with granulation tissue
2.	R. Meher [11]	2003	13/F	-	Left zygomatic arch	Medicinal	
3.	A. Sethi [12]	2006	3/M	-	Left zygomatic bone	Medicinal	
4.	V. Singh [13]	2013	12/F	Father treated for pulmonary TB 1 year back	Right zygomatic bone	Surgical + Medicinal: Curettage through intraoral approach under LA	Complete healing of wound with granulation tissue
5.	N. Uppal [14]	2021	31/M	-	Right zygomatic bone	Surgical + Medicinal: Debridement	Complete healing

## Conclusion

Zygomatic bone tuberculosis is an extremely rare condition that should be considered as a differential diagnosis in patients presenting with facial swellings and discharging sinuses. In cases of extensive damage, secondary infection, or intracranial involvement, a combined approach of anti-tuberculosis therapy and surgical intervention is often required. Proper diagnostic techniques and timely treatment are crucial for ensuring positive outcomes.

## Author contributions

All authors were involved in the conception and design of the study. Dr. Nitin Bhola, Dr. Chetan Gupta, and Dr. Nikhar Wadhvani conducted material preparation, data collection, and analysis. All authors reviewed and approved the final version of the manuscript.

## Consent to publish

The authors confirm that the human research participant gave informed consent for the publication of the images in Fig. 1.

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## CRediT authorship contribution statement

**Nitin Bhola:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Software, Resources, Project administration, Methodology, Investigation, Funding acquisition,

Formal analysis, Data curation, Conceptualization. **Chetan Gupta:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Software, Resources, Project administration, Methodology, Investigation, Funding acquisition, Formal analysis, Data curation, Conceptualization. **Nikhil Wadhvani:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Software, Resources, Project administration, Methodology, Investigation, Funding acquisition, Formal analysis, Data curation, Conceptualization.

### Declaration of Competing Interest

The authors have no relevant financial or non-financial interests to disclose.

### Data Availability

The author confirms that all data generated or analysed during this study are included in this published article.

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