

Primary Tubercular Osteomyelitis Affecting the Mandibular Condyle - A Case Report

Aishwarya Ashok Gupta, Anend A. Jadhav, Nitin D. Bhola, Apoorva S. Mishra, Saurabh Sunil Simre

Department of Oral and Maxillofacial Surgery, Sharad Pawar Dental College and Hospital, Datta Meghe Institute of Medical Sciences (Deemed to be University), Wardha, Maharashtra, India

Abstract

Rationale: Primary orofacial tuberculosis (TB) accounts for <3% of all cases of TB. TB of the mandibular condyle is often misdiagnosed owing to its rarity. **Patient Concerns:** This report presents a 19-year-old female who presented with a painful swelling over the right preauricular region. The radiographic evaluation suggested a diagnosis of suppurative osteomyelitis of the condyle. **Diagnosis:** Clinically, the aetiology of the swelling was considered as infective. The histopathological examination of the resected specimen showed tuberculous granuloma and the polymerase chain reaction came positive. This confirms the diagnosis of tubercular osteomyelitis. Mantoux test and sputum acid-fast bacilli were found to be negative. **Treatment:** The patient was subjected to sequestrectomy with high condylectomy and systemic multidrug antitubercular regimen for 6 months. **Outcomes:** There was a resolution of infection postoperatively with subsequent regeneration of healthy bone. **Take-away Lessons:** Early identification and prompt diagnosis is imperative for better treatment outcomes.

Keywords: Condylectomy, extrapulmonary tuberculosis, mandibular condyle, preauricular region, tubercular osteomyelitis

INTRODUCTION

Tuberculosis (TB) is one of the oldest, daunting microbial infections known to humankind, affecting approximately 20-43% of the world's population.^[1] The orofacial region accounts for approximately 10% incidence of extrapulmonary TB, usually arising from secondary lymphohematogenous dissemination of acid-fast *Mycobacterium tuberculosis*.^[1] Orofacial TB primarily affects the tongue, palate, gingiva and floor of the mouth with rare involvement of jaws. It can be primary or secondary. The primary form can be differentiated from the secondary by its insidious onset, making the diagnosis more challenging. Subjects with secondary form always present with a typical course of TB which is distinctly missing in the primary.

Orofacial tubercular osteomyelitis (TBO) is a rare presentation and poses a diagnostic challenge, with only 3% of TB cases reported in the literature.^[1,2] As the oral cavity is lined by squamous epithelium, it resists the penetration of microbes. The pH of saliva and its contents like anti-microbial enzymes prevents the multiplication and colonisation of the microbes. Apart from haematogenous and lymphatic spread, certain factors help in dissemination of infection such as local

trauma (tooth extraction, fracture of jaws, periodontitis, and periapical pathologies). Other local predisposing factors include poor oral hygiene and hyperkeratotic disorders such as leukoplakia.^[3,4]

The present case report is aimed at highlighting the rare incidence of this “master mimic” and its clinical features and to discuss our clinical experience, thus making the practicing clinicians aware of such rarity.

CASE REPORT

A 19-year-old female presented to the outpatient department with the presence of a single, progressively growing, firm,

Address for correspondence: Dr. Aishwarya Ashok Gupta, Department of Oral and Maxillofacial Surgery, Sharad Pawar Dental College and Hospital, Datta Meghe Institute of Medical Sciences (Deemed to be University), Wardha, Maharashtra, India. E-mail: aishwaryagupta.904@gmail.com

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Figure 1: (a) Frontal view demonstrating swelling over the right side of the face, (b) Lateral view showing swelling over the right preauricular region (arrow)

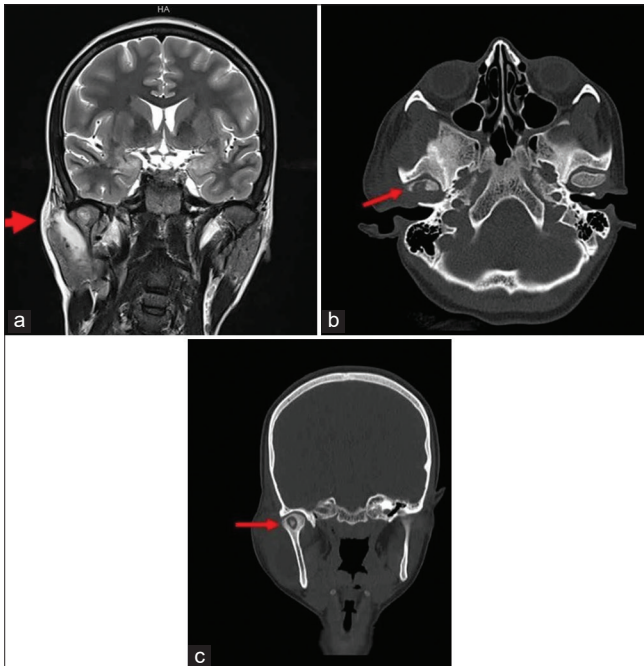


Figure 2: (a) Coronal view of MRI head showing lesion in the right condyle and masseter region. (b) Axial section of CT head demonstrating bony destruction of the right mandibular condyle. (c) Coronal section of CT head showing medullary destruction of the right mandibular condyle.



Figure 3: Chest X-ray of the patient showing no pulmonary lesions

diffuse swelling over the right preauricular region. There has been progressive trismus for eight months which developed insidiously over the past four months. The patient had been on self-prescribed systemic antibiotics for a week with no clinical improvement. The swelling measured 6 cm × 5 cm in the greatest dimension extending anteroposteriorly from the tragal pointer to a line perpendicular to the lateral canthus of the eye and superoinferiorly from the root of the helix to the inferior border of the mandible [Figure 1a and b] with no cutaneous sinuses.

The throbbing and intermittent pain got aggravated on mastication. The swelling had raised temperature and tenderness on palpation. The cervical lymph nodes were not palpable clinically. Detailed medical history and systemic and intraoral examination were unremarkable. The provisional diagnosis of acute exacerbation of chronic arthritis was made. Orthopantomogram revealed an osteolytic lesion of the right mandibular condyle with loss of corticomedullary architecture, multiple cloaca, and sequestrum. T2W1 magnetic resonance imaging of the head demonstrated a hyperintense area over the right masseteric region extending up to the lateral pterygoid muscle with osteolytic configurational changes of the right mandibular condyle [Figure 2a]. Computed tomogram (CT) of the head revealed an osteolytic lesion in the right condyle measuring 6 cm × 4 cm × 2.3 cm, suggestive of suppurative osteomyelitis [Figure 2b and c]. Raised leukocyte, erythrocyte sedimentation rate, and C-reactive protein count were suggestive of underlying chronic infection.

Based on the above findings, a diagnosis of septic arthritis, TBO condyle, and juvenile idiopathic arthritis was made. Mantoux test showed a negative reaction. Chest radiograph [Figure 3] and high-resolution computed tomography of the thorax were unremarkable. Three sputum specimens and fine-needle aspiration cytology (FNAC) of the swelling were found to be smear and culture negative.

Considering the aetiology to be infectious, the patient was subjected to surgical exploration requiring decortication of bone (sequestrectomy) resulting in medullary destruction and cortical perforation followed by placement of corrugated rubber drain [Figure 4a-e]. Histopathology report of the resected specimen revealed extensive inflammatory infiltrate, focal areas of bone necrosis, caseation granuloma, and lymphocytic infiltration with Langerhans' giant cell. This made us to suspect the presence of tuberculous granuloma which is a consistent finding in a diagnosis of TBO [Figure 4f]. A polymerase chain reaction (PCR) of the curetted material showed positivity for TB bacilli.

The patient was prescribed intravenous antibiotics (amoxicillin with clavulanic acid and metronidazole) for 1 week along with a multidrug antitubercular regimen comprising isoniazid (600 mg), rifampicin (450 mg), pyrazinamide (1500 mg), and ethambutol (1200 mg) for 6 months. The patient was advised of monthly follow-up [Figure 5a and b]. The outcome was satisfactory showing

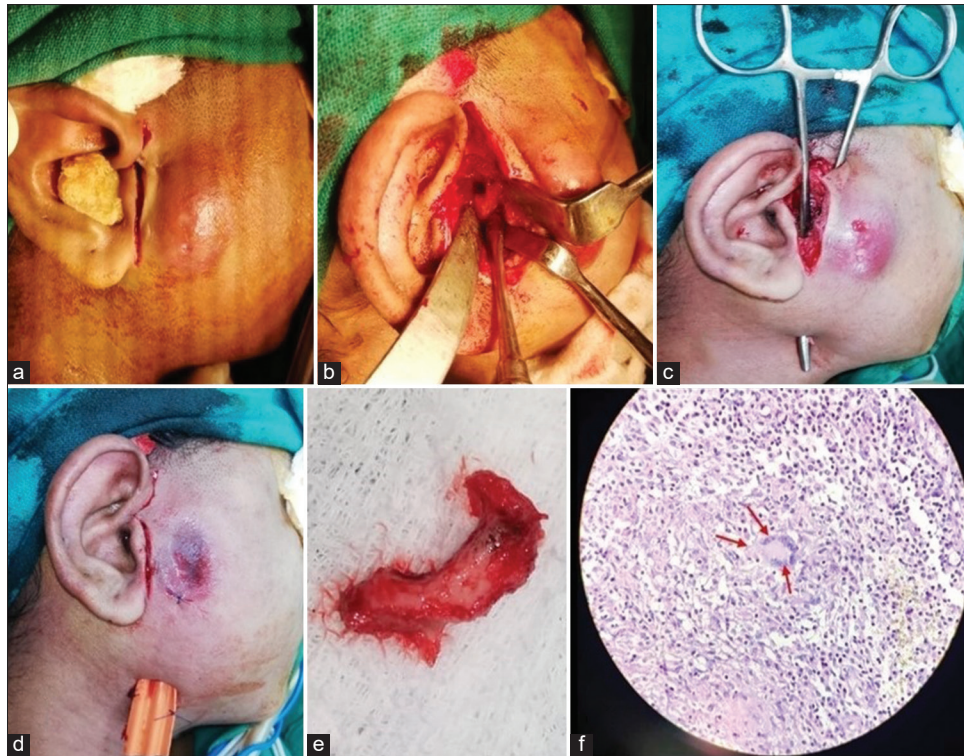


Figure 4: (a) Preauricular incision, (b and c) Surgical exploration and sequestrectomy with high condylectomy, (d) Tissue approximation with placement of corrugated rubber drain, (e) Resected tissue specimen, (f) Histopathology demonstrating Langerhans giant cell, surrounded by lymphocytic infiltration suggestive of chronic granulomatous infection and formation of tuberculous granuloma (Leishman staining, $\times 40$ magnification) (arrows).



Figure 5: (a) Postoperative frontal view after 9-month follow-up, (b) Postoperative lateral view after 9-month follow-up showing healed scar

regeneration of bone at nine months, the surgical site healed completely, and CT (head) demonstrated a healthy remnant bone structure [Figure 6a-c].

DISCUSSION

TB predominantly caused by human and bovine *Mycobacterium bovis* is a multiorgan chronic granulomatous disease that may be a primary or secondary lesion. The human bacillus has a great predilection for all weight-bearing bony joints of the body. Haematogenous dissemination and trauma

are usually considered the major aetiologic factors for TBO of the condyle,^[1,5,6] even though the pathomechanism is multifactorial and the exact aetiology remains unclear.

The features of TBO mandibular condyle constitute preauricular swelling, pain, trismus, pyrexia, pus discharge, and regional lymphadenopathy with/without draining sinuses^[5] as seen in the exhibited case. These are not pathognomonic and can be easily confused with arthritis, osteomyelitis, or a tumour.^[4,6] and the same was observed in our case. The higher propensity of involvement of the posterior ramus and condyle is attributed to haematogenous dissemination owing to the vasculature of the medial pterygoid, masseter muscle from the inferior alveolar artery, and periosteal vessels.

After inoculation, the microorganisms gain access to nutrient vessels and form microthrombi occluding the endosteal and periosteal supply rendering the Haversian system ischaemic and causing the destruction of corticomedullary architecture.^[3] Radiological evidence of ischaemic changes is lacking until three weeks after which an evident cortical sequestrum is seen in the condyle.^[5-7] Removal of this sequestrum is one of the strongest indications for surgical exploration.

The condition has been aptly named “master mimic” by many as it closely resembles other acute inflammatory conditions and tumours.^[3,4,8] The culture requires 4–8-weeks and further 1–2-weeks turnover time for antibiotic sensitivity. Hence, the histopathological diagnosis is better considered to begin an

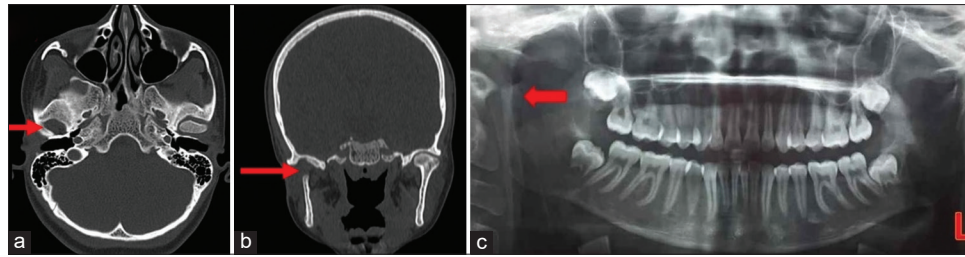


Figure 6: (a) Postoperative axial section of CT head demonstrating surgical defect (arrow), (b) Postoperative coronal section of CT head showing surgical defect (arrow), (c) Postoperative orthopantomograph showing surgical defect after condylectomy (arrow).

appropriate therapy at an early stage of the disease.^[8,9] Except from the conventional acid-fast bacilli (AFB) staining, biopsy, culture and radiological imaging, the newer modalities like bone scintigraphy, cone beam CT, bone PCR and nucleic acid amplification tests can also give confirmatory diagnosis.^[10] In the exhibited case, as the FNAC, Mantoux, and sputum AFB were negative, the diagnosis was confirmed with histopathology and PCR of the curetted material.

Considering the nonspecific presentation, TBO condyle should be considered a differential diagnosis in patients prone to the risk of TB endemic regions and those at risk of HIV.^[3,9,10] Delay in the diagnosis can cause considerable facial deformity and pathological fracture. Antitubercular therapy (ATT) for 6–9 months is advocated by the Revised National TB Control Programme and other anti-TB programs globally.^[8] Surgical debridement of sequestrum, necrotic bone, and drainage of abscess along with ATT is the only surgical modality reported in the literature as performed in this case.^[4,7,8]

The surgical approach used in the present case provides direct access and allows space for condylectomy which may prevent the intracranial spread of infection. Limitations include an evitable scar and facial nerve injuries. The present report highlighted the overall features of TBO by sharing our clinical experience for creating awareness among health professionals.

CONCLUSION

Awareness about orofacial TB is most important in developing countries like India which has the highest prevalence of this infection and the areas of HIV endemicity. It is essential to include TBO as a differential diagnosis of preauricular swellings. Precise diagnosis and prompt treatment with antitubercular drugs are the mainstays of management.

Declaration of patient consent

The author certifies that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient's demographic details will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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