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Tuberculous Osteomyelitis of the Mandible: An Unusual Clinical Presentation

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ABSTRACT

Tuberculosis can affect any part of the body with varying severity; that in the cranio-facial bones is relatively rare. When compared to pyogenic infections and neoplasms affecting the mandible, tuberculous osteomyelitis of the mandible is extremely rare. We report a case of tuberculous osteomyelitis in a 25-year-old female patient who came to us with complaint of pus discharge intraorally from lower left canine region which later proved to be tuberculous osteomyelitis with no primary focus and which responded well to anti-tubercular treatment. One should always suspect mycobacterial infection when dealing with chronic maxillofacial infections not responding to the usual antibiotic course and local debridement.

Key words: Tuberculous osteomyelitis, mandible, GeneXpert, young adult

INTRODUCTION

Osteomyelitis is nothing but an inflammation of the bone marrow that has a tendency to progress. This differentiates the dentoalveolar abscess, "dry socket" and "osteitis," seen in infected fractures from osteomyelitis. It involves adjacent cortical plates and often periosteal tissues. Osteomyelitis of the jaws is predominantly a disease of the mandible, whereas the maxilla by virtue of its vascularity and thin cortical plates is less frequently involved.[1]

Diminished local and systemic host defences can contribute to the emergence of the disease as well as its clinical course. Osteomyelitis has been found to have association with multiple systemic diseases including diabetes, malignancies, malnutrition, acquired immunodeficiency syndrome etc. Medications like steroids, chemotherapeutic agents, and bisphosphonates alter the vascular dynamics of bone and hence predispose to osteomyelitis. Local conditions adversely affecting the blood supply can also predispose the host to bony infection.

Depending on the clinical presentation, suppurative osteomyelitis may typically be acute or chronic. Acute osteomyelitis which is characterised by inflammation and suppuration within the marrow space needs to be suspected when there is onset of paresthesia of the inferior alveolar nerve, despite the absence of radiological signs. This drifts into the chronic phase when inadequately treated, or the host defence attains partial control or

when the pus within the marrow spaces finds its way out by cortical perforation and sinus tract formation.

Other forms of non-suppurative osteomyelitis include osteoradionecrosis (ORN), bisphosphonate related osteonecrosis of the jaws (BRONJ), Garre` osteomyelitis, chronic recurrent multifocal osteomyelitis of children, and chronic sclerosing osteomyelitis.

Tuberculosis (TB) is common in developing countries. Tuberculous osteomyelitis is a rare chronic non-suppurative form of osteomyelitis that occurs more often in young individuals and in most cases is detected in the late stage of the disease. The most common sites involved are dorsal and lumbar vertebrae and epiphysis and diaphysis of long bones. Flat bones, including those of skull and mandible are rarely affected. Occurrence of tuberculous osteomyelitis in the jaw is very low. It rarely arouses clinical suspicion because of such low incidence and atypical presentation.

In the clinical practice of oral and maxillofacial surgery it presents as an enigma for the clinician in terms of diagnosis and treatment. Delay in recognition of the infection may result in a protracted course of treatment and increased surgical morbidity.[1]

CASE REPORT

A young 25-year-old female was referred by a private practitioner to the department of Oral and Maxillofacial Surgery at MGM Dental College and Hospital with complaint of swelling and pus draining from lower labial vestibule in relation to the lower left canine.

Patient was vitally and systemically stable. On routine investigations, she was found to have microcytic hypochromic anemia. Apart from that, there was no other complaint. She had over-retained deciduous lower left canine, grade II mobile 73, grade I mobile 41 31 32 34 and missing 33. The intra-oral swelling was tender with erythematous overlying mucosa and draining sinus tract. There was a non-tender palpable submandibular lymph node on the same side. Routine orthopantomograph (figure 3) showed an ill-defined radiolucent lesion in the subapical region extending from 43 till 36 with the presence of an impacted canine. The apices of 32, 73 and 34 were involved and gave no response on electrical pulp testing. The other teeth in the vicinity were found to be vital. A cone beam computed tomography was done (figure 2) to view the precise extent and involvement of the lesion. It showed buccal and lingual plate perforation. A differential diagnosis of osteomyelitis, infected radicular cyst, chronic dentoalveolar abscess, dentigerous cyst and odontogenic keratocyst was given.
Biopsy was done where the H & E stained soft tissue sections showed presence of granulomatous inflammation with well-formed clusters of epithelioid histiocytes. Mantoux test was done that came out negative. Chest x-ray and USG chest were also done that suggested right sided non-tappable minimal pleural effusion.

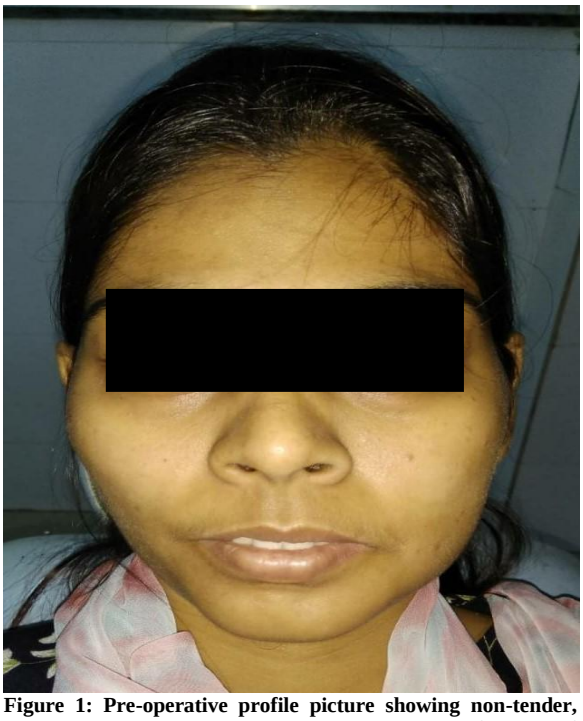

Figure 1: Pre-operative profile picture showing non-tender, non-erythematous, localized swelling on the left side of mandibular parasymphysis region

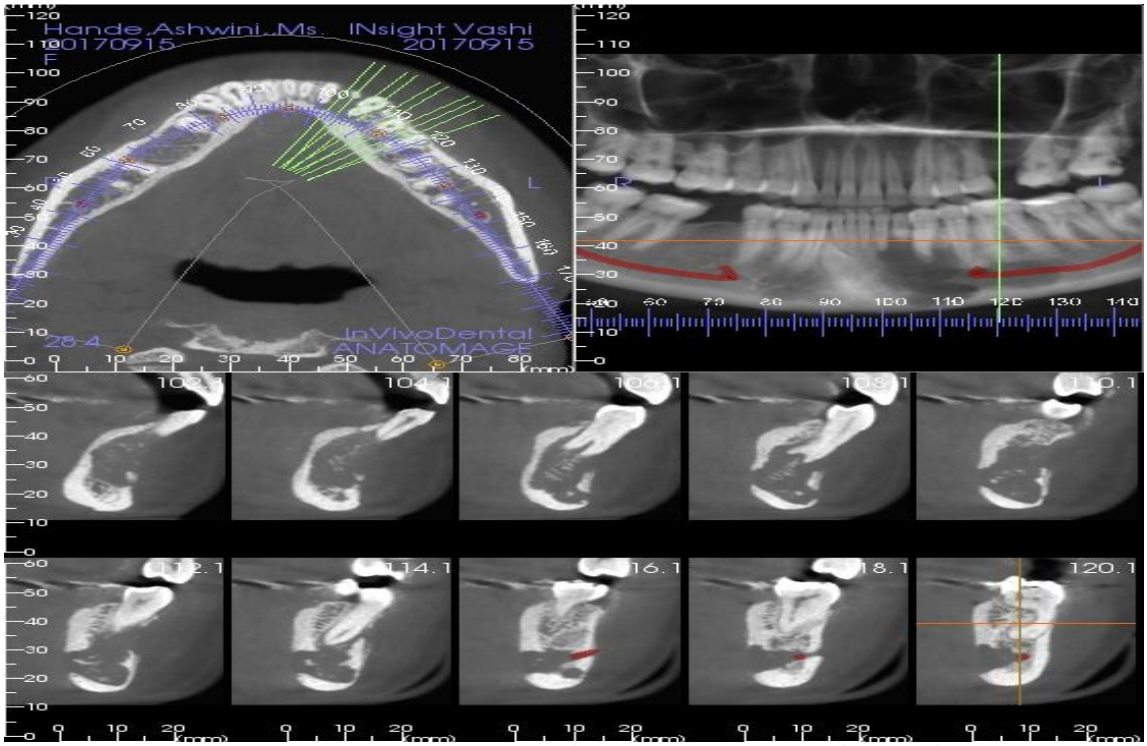

Figure 2: Pre-operative CBCT sections showing involvement of mandibular bone with perforation of both lingual and buccal cortices

Since the biopsy report was inconclusive and the lesion was well confined, a complete curettage as well as extraction with impacted canine, deciduous canine and first premolar was done and the tissue was sent for histopathology. It was found to be chronic inflammatory lesion (granulomatous) in nature. The patient was asymptomatic thereafter until 2 months later when she again presented with a similar swelling in the same region. This time pus was aspirated, caseous white material was curetted and the sample was sent for gene expert. Xpert MTB-RIF Assay G4 was performed and reported as MTB DETECTED LOW (low levels of mycobacterium tuberculosis) with Rif RESISTANCE NOT DETECTED (no resistance to Rifampicin). Sample which was cultured showed Streptococcus Pneumoniae. Patient was referred to Chest-TB Physician. She was put on AKT regime for 6 months and kept under close follow-up

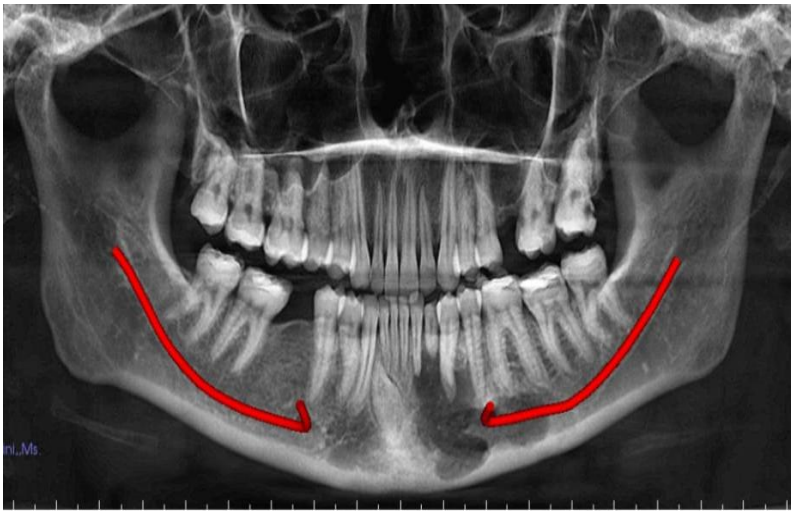

Figure 3: Pre-operative radiograph shows multiple radiolucencies within the mandible extending from 36 till 43 with presence of impacted canine

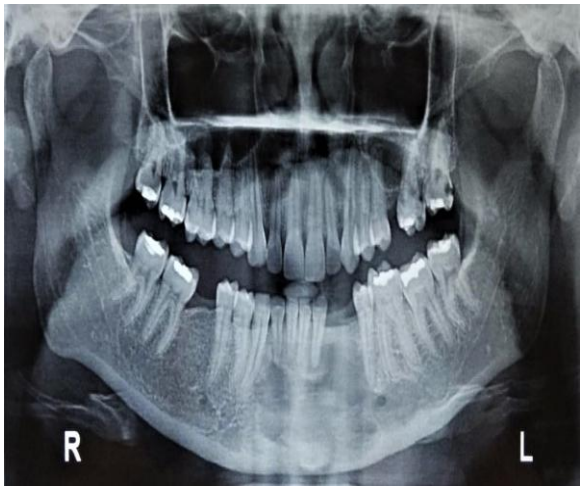

Figure 4: 1-month post-operative radiograph done after the second excisional biopsy shows healing in the same region with mixed radiopacity and radiolucency

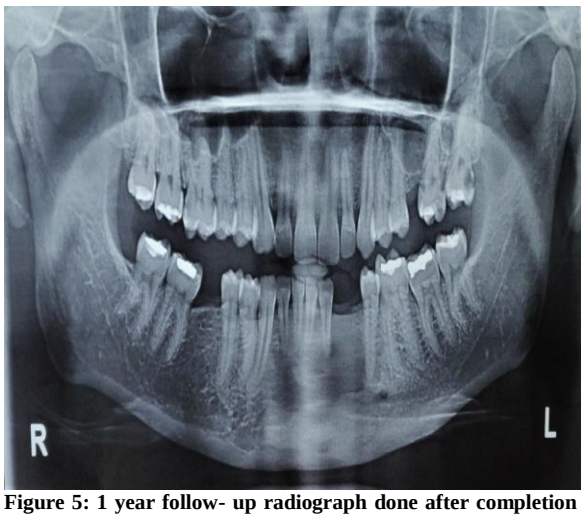

Figure 5: 1 year follow- up radiograph done after completion of the AKT regime shows adequate bone formation in the region of the lesion

Figure 6: 2-year follow-up clinical picture shows good healing and no signs of infection

DISCUSSION

Tuberculosis (TB) ranks first in the top 10 causes of death worldwide. Tuberculosis is estimated to affect 1.7 billion individuals worldwide, with eight to ten million new cases and 1.7 million deaths each year. EPTB (Extra-Pulmonary TB) sums up for about 15% to 20 % of all cases of TB. The annual global incidence of EPTB has been increasing in the last decade due to the changing TB control practices, spread of HIV (human immunodeficiency virus), the population growth and the cure of infectious cases of TB might have resulted in a relative rise of annual EPTB case detection[2].

Of the reported cases of mandibular tuberculosis, most are secondary to focus elsewhere in the body. As such primary tuberculosis of the mandible is rare occurrence. Tuberculosis of the mandible apparently affects both sexes, with male preponderance. According to Chapotel, more than 60% cases of tuberculous osteomyelitis of the mandible are seen in patients less than 15 years of age; but it can be seen in old age as well[3].

Tuberculous infection of oral tissues can be primary or secondary. Primary lesions develop when tuberculosis bacilli are directly inoculated into the oral tissues like gingiva, tooth extraction sockets and buccal folds, of a person who has not acquired immunity to the disease. Infection of oral tissues can result from either hematogenous or lymphatic spread or from autoinoculation by infected sputum and direct extensions from neighbouring structures[4].

Oral lesions of TB, although uncommon, have certain manifestations. Commonly seen signs of tuberculous infection of the oral cavity include ulcers, granulomas, pain and swelling, mobile teeth, and displacement of tooth buds[5].

Mandibular tuberculosis presents as a multifocal lesion elsewhere in body, involving other bones and lungs. It causes slow bone necrosis and may progress to involve the entire mandible. Radiographically, bone destruction appears as blurring of the trabeculae with irregular radiolucency and cortical erosion which has

little tendency to repair. Eventually bone is replaced by soft trabecular granulation tissue. Caseation appears followed by softening and liquefaction. A soft periosteal abscess then forms presenting as a painless soft swelling. This cold abscess may burst either intra-orally or extra-orally later forming single or multiple sinuses. This might result in pathological fracture of the mandible and sequestration of same[3].

As was evident in the present case, diagnosis of mandibular tuberculous osteomyelitis is significantly difficult since there are no specific signs which are pathognomic of the infection. The only manifestation may be a localized swelling of the jaw which has high chances of being misdiagnosed as a pyogenic abscess or if sinuses are present, may be confused with other granulomatous diseases like actinomycosis.

Diagnosis of extra-pulmonary TB (EPTB) was particularly challenging as the number of Mycobacterium tuberculosis (MTB) bacilli present at the site of involvement was low. Tissue microscopy only revealed nonspecific chronic inflammatory changes and special staining was not useful. Isolation of *M. tuberculosis* from clinical samples by culture is the "gold standard" for a definitive diagnosis of EPTB. However the feasibility and reliance on this report for definitive diagnosis somewhat delayed recognition. Repeated tissue screening and cultures failed to throw light on the diagnosis in the involved case.
In such circumstances, newer diagnostic direct methods like the Xpert MTB/RIF, BACTEC culture system, Immunohistochemistry and Immunocyto-chemistry & indirect methods like Interferon-release assay ensure high sensitivity and specificity. The flip side is that the BACTEC system is expensive and has associated radio-active hazards while immunohistochemistry and immunocyto-chemistry methods cannot identify viable bacteria nor make drug sensitivity feasible[6].

With the advent of the Xpert MTB/RIF automated molecular assay (Cepheid, CA, USA) for quick diagnosis of TB and detection of Rifampicin resistance, which is a marker of MDR-TB, a landmark milestone has been achieved in TB research. This multifunctional diagnostic assay is an automated, closed system that performs real-time PCR and can be used by operators with minimal technical expertise, facilitating diagnosis of TB and simultaneous assessment of rifampicin resistance to be completed within 2 h[7]. The assay was developed, optimized, assessed, and endorsed specifically for the detection of pulmonary TB using sputum. More recently, however, assessments of the assay have extended to various extra-pulmonary clinical samples. The reported sensitivity of the assay for EPTB are highly heterogeneous, ranging from 25% to 96·6%[6].

In the case presented, the gold standard testing that has been considered conventionally did not yield any result and delayed the diagnosis. The advent of newer diagnostic methods, like GeneXpert as used here helped fast track the diagnosis and the treatment.

In view of the current case scenario, it would be right to say that one should not rely solely on the conventional methods but put to use the newer methods as adjuncts aiming at faster diagnosis and thereby reducing delay in the management.

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