

Case Report

Orofacial tuberculosis: A rare case report and a review

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Abstract

Tuberculosis is the leading cause of death worldwide with India leading the count (World Health Organization report 2016). We present a case of right submandibular swelling and tuberculous periapical granuloma associated with carious mandibular 2nd molar adjacent to partially impacted third molar in a 35-year-old female patient. Histopathology report of periapical specimen revealed tuberculous granulomatous pathology that correlated with the histopathology report of submandibular lymph node on the same side of face. The lesion healed uneventfully with expert medical consultation. The patient was followed up for 13 months.

Keywords: Caseating granuloma, langhans multinucleated giant cells, periapical granuloma, tubercular lymphadenitis, tuberculoma, tuberculous granulomatous pathology

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INTRODUCTION

Tuberculosis (TB) is one of the top 10 causes of death worldwide. Six countries account for 60% of the total, with India leading the count. TB is caused by bacteria (*Mycobacterium tuberculosis*) that most often affect the lungs. About one-third of the world's population has latent TB.^[1] When a person develops active TB disease, the symptoms (such as cough, fever, night sweats, or weight loss) may be mild for many months. This can lead to delays in seeking care, and results in transmission of the bacteria to others.^[1] Several studies report that TB is less common in females than in males.^[2,3] In the present case, an afebrile middle-aged female presented with tuberculoma associated with 47 and buccally draining abscess associated with submandibular swelling.

CASE REPORT

A 35-year-old female patient was referred to the Department of Oral and Maxillofacial Surgery for

extraction of grossly carious 47 and partially impacted 48. She had associated history of submandibular swelling on the right side from the past 4 months that did not resolve after taking antibiotics and analgesics. She revealed no other contributory medical history. There was no history of fever or weight loss. On examination, she had approximately 3 cm × 4 cm, hard, nontender, nonfluctuant fixed swelling on the right submandibular region. Intraoral examination revealed buccally draining abscess with relation to carious 47 and partially impacted 48. Preoperative IOPA and panoramic radiograph [Figure 1a and b] shows the presence of periapical pathology with respect to 47 and impacted 48. She was afebrile and her vitals were stable. She was provisionally diagnosed with chronic dentoalveolar abscess in relation to 47 adjacent to impacted 48 and also associated with right submandibular lymphadenitis that did not resolve with oral antibiotics and anti-inflammatory drugs. Tooth extractions along with periapical curettage

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were done under strict aseptic condition. Periapical diseased tissue was sent for histopathological examination. Histopathological findings of the intraoral periapical soft tissue specimen obtained with respect to 47 and 48 region revealed tuberculous granulomatous pathology and was advised to rule out TB by the Department of Oral and Maxillofacial Pathology [Figure 2]. This is not usually seen in intraoral lesions but may be an important component in lymph node involvement and the lung.^[4]

She was referred for expert medical opinion. Fine-needle aspiration cytology (FNAC) of the right submandibular lymph node revealed tuberculous granulomatous pathology that correlated with the histopathological findings of periapical specimen. Zeil neilson stain for acid-fast Bacilli (AFB) was negative. Chest X-ray revealed the presence of foci of pulmonary lesion in left lung field as shown in [Figure 3a]. Based on these findings, her antitubercular therapy regime AK4 followed by the course of AK3 was started and continued for 6 months. The patient was recalled for checkup. Her postoperative panoramic radiograph [Figure 4] and chest X-ray [Figure 3b] at 13th month follow-up period is shown.

Intraoral healing [Figure 5b] was uneventful. The right submandibular scarring was also observed [Figure 5a].

DISCUSSION

The World Health Organization estimates that in India approximately 300,000 people die from TB each year, while the Global Burden of Disease study 2013 estimates >0.5 million deaths/year.^[5] A large number of dentists and consultants have limited experience with the TB of the upper aero digestive tract since its oral lesions are nonspecific in its clinical presentation and are often ignored in the differential diagnosis. This is very common in cases where oral lesions are present before the systemic symptoms become apparent.^[6]

Shafer reported that males are more affected than females in India contrary to our case. Several reviews have discussed the possibility of under notification of women due to greater difficulties in gaining access to clinics and in obtaining a timely diagnosis and treatment, particularly in developing countries.^[2] Even poor quality of sputum samples collected

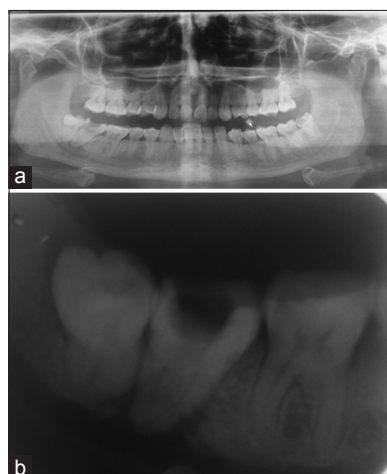


Figure 1: (a) Preoperative orthopantograph showing periapical pathology with respect to carious 47 and impacted 48. (b) Preoperative intraoral periapical showing periapical pathology with respect to carious 47 and impacted 48

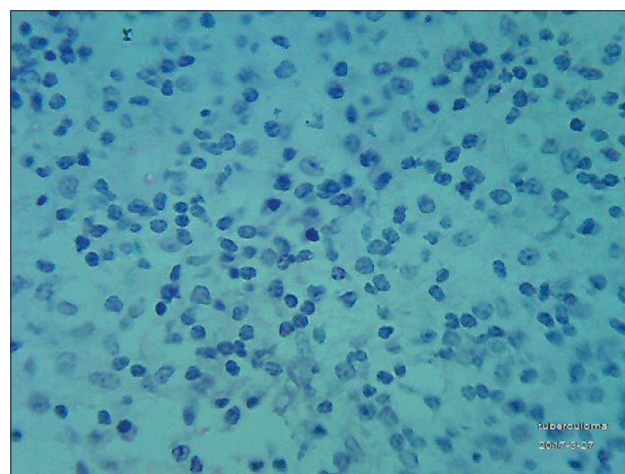


Figure 2: Histopathology picture of intraoral soft tissue specimen as seen through microscope on glass slide stained with hematoxylin (blue) and eosin (red) dyes. Caseating granuloma along with numerous randomly distributed multinucleated langhans type giant cells in this picture were found in the periapical curettage specimen of a patient

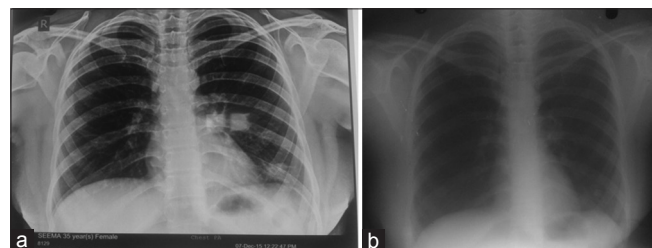


Figure 3: (a) Perioperative chest X-ray showing: a small patch of infiltration is seen in the left lower zone. Right lung field is clear. (b) Postoperative chest X-ray after 13 months

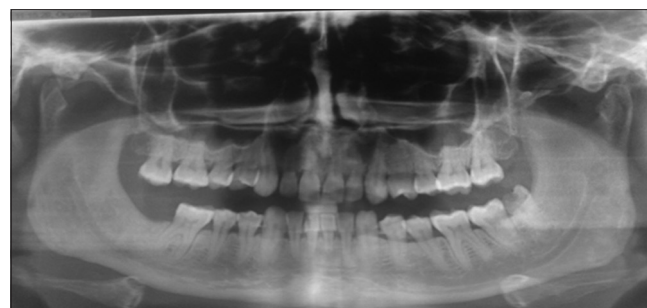


Figure 4: Thirteenth-month postoperative panoramic radiograph showing missing 47, 48 with no pathology on the right side of mandible



Figure 5: (a) Postoperative view of submandibular region showing scar after 13 months. (b) Intraoral picture showing healthy healing

from women in some regions, may influence the sex bias observed in patients with TB.^[3] However, in concordance to our case Nidhi *et al.* in their study on FNAC in three hundred and eighteen consecutive superficial lymph nodes, clinically suspected to be tuberculous in a tertiary care center reported female preponderance, 75% of the patients in second and fourth decade of life and cervical region was most affected with solitary tuberculous lymphadenitis.^[7]

TB presents in many forms and is known to affect almost any part of the human body. Extrapulmonary sites, such as those involving the gastrointestinal system, central nervous system, skin, lymph nodes, or oral cavity, may also be involved as a primary or secondary lesion.^[8] Our case shows chronic submandibular swelling and periapical tuberculoma.^[2] Andrade and Mhatre in their case on Orofacial TB a 16-year experience with 46 cases noted that angle of mandible was the most common site.^[8]

Human strains of *M. tuberculosis* are responsible for many cases of TB but bovine strain of may also produce illness through ingestion of unpasteurized cow's milk. Rarely, atypical or opportunistic bacteria can cause pulmonary or generalized infection in immunocompromised individuals.^[2] There may be local factors such as poor oral hygiene, local trauma, preexisting lesions such as periapical granuloma, cysts, and abscesses, and periodontitis.^[8] In the present case spread to periapical region was through odontogenic infection and pericoronitis. A systemic predisposing factor, such as altered host resistance from immunosuppression or nutritional deficiencies, increases the susceptibility of a patient to a tuberculous infection.^[8]

Even on histological examination, we see a granulomatous lesion, we ought to consider other entities such as sarcoid, Crohn's disease, cat scratch disease, foreign body reaction, and tertiary syphilis.^[7,9]

After referral to a specialist 10 days after extraction of teeth, report of the periapical specimen correlated with the FNAC report (done at referral center) of the right

submandibular lymphadenitis. Her chest X-ray [Figure 3] also showed positive findings for TB. In 1994, Molinari and Coltone^[10] reported the use of FNAC for vertebral TB. Diagnostic accuracy as high as 100% in tuberculous lymphadenopathy cases has been reported by FNAC in comparison to excisional biopsy.^[7]

Ten point protocol developed by Andrade *et al.* for evaluation and treatment of orofacial tuberculosis is as follows:^[7]

1. A complete history should be obtained from the patient to rule out active pulmonary TB or a family history of TB, and a clinical examination should be performed to rule out odontogenic sources of infection
2. Chest radiographs, orthopantomograms, and computed tomographic scans should be obtained. Full-body scintigraphy should be performed to rule out skeletal foci of infection in other parts of the body
3. Purified protein derivative (i.e., Mantoux test) and erythrocyte sedimentation rate should be determined
4. Polymerase chain reaction for immunoglobulin G and immunoglobulin M antibodies for M TB should be performed
5. Sputum samples (3 samples) should be scrutinized for AFB
6. For "lumpy jaw" (incision and drainage were not performed), aspiration should be performed and the aspirate should be assessed by histopathology (Ziehl-Neelsen stain) and culture using Bactec (Becton Dickinson Pvt Ltd., Haryana, India)
7. For extra- or intraoral draining of sinuses and unhealed extraction sockets or deep-seated bony lesions, curettage, or incisional biopsy should be performed and assessed by histopathology (Ziehl-Neelsen stain) and culture with BACTEC
8. Only those patients with a diagnosis positive for TB by histopathology with or without positive culture studies should be started on anti-TB therapy under a directly observed treatment strategy
9. Long-term follow-up is mandatory.

CONCLUSION

It is concluded that complete history, examination and investigations are mandatory for the differential diagnosis of orofacial TB. A strict aseptic protocol must be followed for the management of orofacial TB. Long-term follow-up is necessary.

Clinical significance Although middle aged males are more commonly affected by TB, there are several factors that influence sex bias. Our case presents Orofacial TB in a middle aged female patient, so

more studies are required to assess females affected by orofacial TB. For extra or intraoral draining of sinuses and unhealed extraction sockets or deep seated bony lesions, curettage or incisional biopsy should be performed and assessed by histopathology (Ziehl Neilsen stain) and culture.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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