

Tuberculous osteomyelitis of the maxilla: A rarest of rare case report

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ABSTRACT

Tuberculosis is a chronic granulomatous systemic infectious disease caused by *Mycobacterium tuberculosis*. The oral lesions found in tuberculosis are relatively rare and may present as ulcers, erythematous patches, indurated lesions, nodules or as bony jaw lesions. Oral tubercular lesions sometimes present a confusing clinical presentation and can be overlooked. Hence, we document a case of tuberculous osteomyelitis of the maxilla in a 19-year-old female patient, who was initially treated for multiple periodontal dental abscesses, which later proved to be tubercular osteomyelitis of the maxilla. Although it is a rare occurrence, the differential diagnosis of tuberculous osteomyelitis must always be considered when it fails to respond to routine therapy.

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INTRODUCTION

Tuberculosis is a chronic granulomatous systemic infectious disease prevalent worldwide, which can affect any part of the body, including the oral cavity. It is usually caused by inhalation of infected airborne droplets containing the *M. tuberculosis bacillus*, which is an aerobic, slender, non-motile, non-encapsulated, non-sporing, rod-shaped organism, ranging from 2 to 5 µm.^[1,2]

Oral tuberculous lesions are relatively rare. Oral manifestations in tuberculosis occur in 3% of the cases involving long-standing pulmonary or systemic infection.^[3] Oral lesions may present as ulcers, erythematous patches, indurated lesions, nodules or as bony jaw lesions.^[4] Bony tuberculosis of the jaw is an uncommon form of extrapulmonary tuberculosis and is very rare. Tuberculosis of the jaw is more common in

children below the age of 18 years.^[5] Tubercular infections can be recognized as primary and secondary. Primary lesions develop when the tuberculosis bacilli are directly inoculated into the oral tissues of a person who has not acquired immunity to the disease. Secondary infection of the oral tissues can result from either a hematogenous or lymphatic spread.^[6,7] The involvement of the maxilla by tuberculous infection is the rarest of rare, although tuberculous osteomyelitis of the mandible has been reported and is more common than that in the maxilla, as it contains a lesser amount cancellous bone.^[8,9] The oral tubercular lesions can sometimes present with a confusing clinical presentation. Hence, we document a case of tuberculous osteomyelitis of the maxilla in a 19-year-old female, who was initially suspected for multiple periodontal abscesses, which later proved to be tubercular osteomyelitis of the maxilla.

CASE REPORT

A 19-year-old female patient reported to the Department with a complaint of mobile teeth along with a history of pus discharge since three months. The history revealed that the patient had a painful swelling in the upper front region of the face three months back, which later subsided due to formation of multiple draining sinuses. The medical history revealed that the patient had

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generalized weakness, weight loss, and episodic fever for the last one month. On general examination, the patient was poorly nourished and thin built. On extra oral examination a diffuse swelling was present in the middle third of the upper left and right side of the face, which extended from the right to the left mid pupillary line mediolaterally and from the ala tragus line, 4 cm inferior to the infraorbital margin. Superoinferiorly the swelling was slightly tender on palpation [Figure 1]. On intraoral examination, multiple draining sinuses were present with reference to the 23, 26 and 14, 15 extraction socket region; there was discharge of pus from all the sinuses and from the extraction socket [Figures 2-4]. On digital palpation (using index finger and thumb) a very unusual finding was found, the maxillary teeth and maxillary arch in their entirety were movable, like a paper-thin bone [Video 1]. There was no carious tooth associated with the sinuses, so on the basis of the above-mentioned history and clinical presentation a provisional diagnosis of multiple periodontal abscesses was made. Routine hematological, radiological, and histopathological investigations were carried out. On the hematological

investigation, the erythrocyte sedimentation rate (ESR) was seen to be very high, which indicated a deep-seated infection. On a panoramic examination, diffuse bone loss in the maxilla, involving all the maxillary teeth, was seen [Figure 5]. A sputum examination for acid fast bacilli (AFB) was inconclusive. A chest x-ray showed that the bronchovascular markings were accentuated and in the left upper zone Pulmonary Koch's was found [Figure 6]. A Montoux test was advised, which came positive (>10 mm). A cytological smear taken from the sinus drainage was stained with Ziehl-Neelsen and Periodic acid-Schiff (PAS), but revealed only polymorphonuclear leucocytes and few lymphocytes. Fine needle aspiration cytology (FNAC) showed no granulomatous lesion. A serological examination for *M. tuberculosis* antibody was performed, which showed a raised level of IgG and IgM antibodies. Finally, an incisional biopsy, which is considered the gold standard, was performed in the extraction socket 14, 15 region [Figure 7]. It showed granulomas of varying sizes and shapes, consisting of central Langhan's giant



Figure 1: Profile picture of the patient



Figure 2: Intraoral view showing multiple draining sinuses



Figure 3: Intraoral sinuses with reference to the 23, 25 region



Figure 4: Draining sinuses with reference to the 14, 15 extraction socket

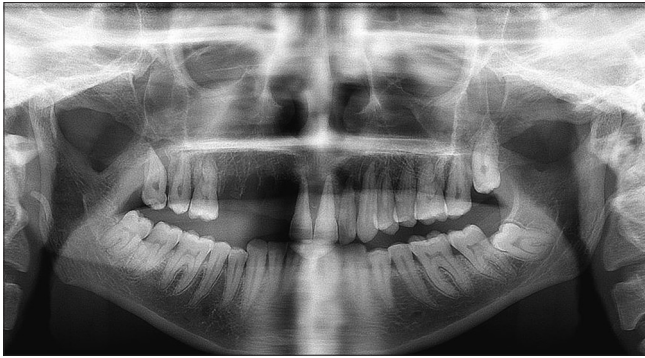


Figure 5: Panoramic x-ray showing diffuse bone loss in the maxilla

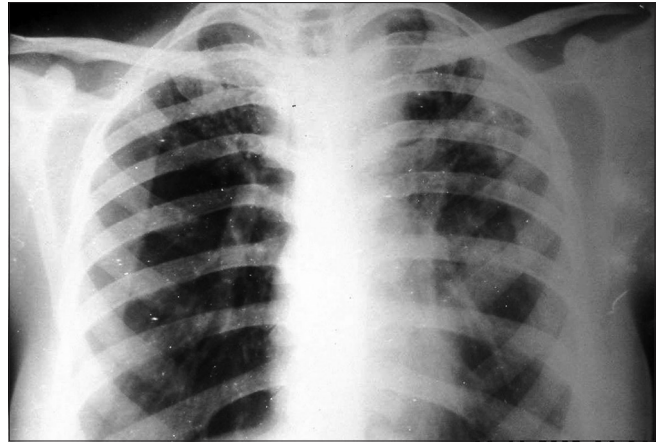


Figure 6: Chest x-ray shows bronchovascular markings are accentuated, with left upper zone Pulmonary Koch's

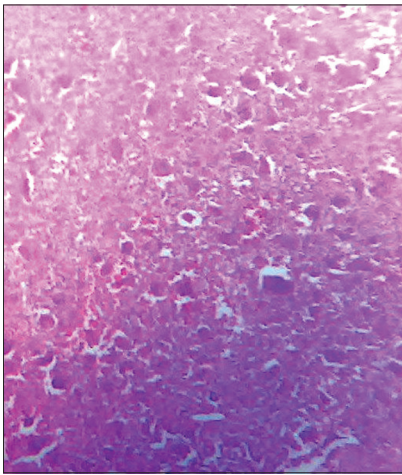


Figure 7: Photomicrograph showing multinucleated giant cells



Figure 8: Incisional biopsy done with reference to the 15 region

cells, epithelioid cells, surrounded by lymphocytes, and few plasma cells [Figure 8]. The above-mentioned features are consistent with a tuberculous granuloma, a chronic granulomatous lesion. Therefore, a final diagnosis of pulmonary tuberculosis as a primary lesion, with a secondary lesion in the maxilla, was made. The standard anti-tubercular therapy for bone tuberculosis was started. At the four-week follow up, a marked improvement in the overall health was noticed, with an overall reduction in the mobility of the maxillary bone and teeth [Figure 9 and Video 2].

DISCUSSION

In spite of the dramatic improvement in public health, tuberculosis still remains a dreaded disease in today's world. A majority of the cases belong to the developing countries, with the highest incidence in the Indian and African subcontinent.^[10,11] The epidemiology of tuberculosis differs considerably with ethnicity, age, and socioeconomic group.^[11,12] Primary oral tuberculosis is rare because of various barriers present in the oral cavity, such as, intact oral mucosa, salivary enzymes, tissue antibodies, and oral saprophytes. Any breach in these barriers can lead to infection by the *tubercle bacilli*.^[2,10]

Tuberculous osteomyelitis is even rarer and constitutes less than 2% of skeletal TB. Jaw involvement of the maxillary bone by TB infection is extremely rare.^[8,9] The radiographic appearance of TB of the jaws or alveolar bone is similar to that caused by other pyogenic organisms. It usually appears as a blurring of the trabecular pattern, with irregular areas of radiolucency.^[13]

Generally the diagnosis of tuberculosis has been made on the basis of clinical findings and radiographs, along with sputum examination showing AFB bacilli. In recent times, more advanced investigations like a DNA probe and polymerase chain reaction assays allow a more sensitive and rapid diagnosis.^[13] The smears and culture for AFB from the oral lesions and the sputum have been negative in our case, which can be due to many reasons. High immunity of the patient, small numbers of *tubercle bacilli* in the oral lesion or a previous history of prolonged antibiotic consumption.^[14] In our case, the very rarest of rare finding was noticed, the entire maxillary arch was moving like a paper-thin bone and drastic reduction was seen in the mobility after initiating anti-tuberculosis treatment.



Figure 9: Post-treatment picture showing complete healing of the sinuses

CONCLUSION

Tuberculous osteomyelitis of the maxilla is an extremely rare condition of the oral cavity. It can present in an unusual form in the oral cavity and can be misdiagnosed. With the recent reversal in the incidence of tuberculosis; it must always be included in the differential diagnosis. Concurrent pulmonary lesions should also alert the oral physician to locate a primary site of the disease in the body, before diagnosis of oral tuberculosis is considered.

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