Primary Tuberculous Osteomyelitis of Maxilla: A Rare Entity
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Abstract
Granulomatous lesions represent a unique form of chronic inflammatory response. Tuberculosis is the most common granulomatous lesion. In spite of taking several measures to eradicate tuberculosis, it still persists in the developing countries. Its incidence has been increased in patients with HIV infection. This primarily affects lungs but also occur in the intestine, meninges, bones, joints, lymph nodes, skin and rarely oral cavity. Primary oral tuberculosis is diagnostic challenge to the clinicians, as it presents with non-specific signs and symptoms. One such case of primary oral tuberculous osteomyelitis affecting the maxilla is reported here.

Keywords: Tuberculosis, Osteomyelitis, Maxilla.

Introduction
Granulomatous inflammation affecting oral soft and hard tissues are rare. Tuberculosis is a chronic infectious granulomatous bacterial disease caused by mycobacterium tuberculosis. Mycobacterium tuberculosis is a Gram positive, acid-fast bacillus. It was first described by Robert Koch in 1882. It commonly affects lungs and other structures such as lymph nodes, intestine, meninges, skin and oral cavity. It presents as primary and secondary form. Primary form is by direct inoculation of organisms into the oral tissues whereas secondary form through lymphatic and haematogenous spread by the infected sputum. In the head and neck region, tuberculosis mainly presents as cervical lymphadenitis. However 0.8-3.5% of pulmonary tuberculosis patient present with oral manifestation. Extra pulmonary tuberculosis manifestations may be identified in the oral cavity but primary oral tuberculosis is extremely rare. Dorsal surface of the tongue is most common site for oral tuberculosis, followed by the lower jaw, buccal mucosa and the lips. Intra osseous tuberculosis is extremely unusual. Oral tuberculosis as a Primary lesion is diagnostic challenge to the clinicians, due to its presentation with non-specific signs and symptoms. Hence, we are presenting a case of primary oral tuberculous osteomyelitis affecting the maxilla.

Case Report
A 39 years old female patient reported with swelling and pain in the upper left region since 5 months. Initially swelling was small to begin with and the swelling slowly progressed to the present size of 3×4 cm and it was associated with pain and pus discharge. Pain was moderate in nature and localized. Past medical history was not significant. Patient had undergone extraction of Grade II mobile teeth in relation to 26 and 27 one month back. Left submandibular lymph nodes were palpable and the aspiration was negative.

On extra oral examination, there was a diffuse swelling along the left upper jaw extending from philtrum to 3cm lateral to the ala of the nose. Intra orally, the swelling extended from the left central incisors to the left 1st molar region expanding both buccal and palatal cortical plates. Root canal treatment was done in relation to 25. Grade I mobility of teeth were noticed in relation to 21, 22, 23, 24 and 25. Orthopantamogram radiograph (OPG) showed ill-defined radiolucency in relation to 21, 22, 23, 24, 25 extending to the posterior border of maxilla. Computed Tomography (CT) showed absence of bone separating left maxillary molar tooth socket from left maxillary sinus with loss of continuity of palatal cortical plate (Figure 1 & 2). Areas of permeative radiolucencies with bone expansion and areas of lucency were seen in the left maxillary alveolar process and the roots of left maxillary teeth from incisors to molars. Cortical bone breach was seen separating the maxillary alveolus from the nasal cavity. A provisional diagnosis of osteomyelitis was given.

An Incisional biopsy was performed in the region of 24 and 25. The tissue was formalin fixed, processed, sectioned and stained. The
given hematoxylin and eosin stained section showed hyper cellular fibro vascular connective tissue with granuloma formation (Figure 3). Numerous chronic inflammatory cells predominately of lymphocytes and plasma cells were evident throughout the connective tissue. Multi nucleated Langhan’s type of giant cells with 8-10 nuclei arranged in horse shoe shaped were evident in the connective tissue (Figure 4). At focal areas, resorbing dead bone was evident. The histopathological features were suggestive of chronic granulomatous lesion and were consistent with tubercular osteomyelitis of maxilla.

Figure 1: The computed tomography images showed absence of bone separating left maxillary molar socket from left sinus (a) with loss of continuity of palatal cortical plate (b). The photomicrograph of hematoxylin and eosin stained section under scanner power view (c) shows granuloma formation within the hyper cellular stroma, whereas under low power view (d) multinucleated Langhans type of giant cells with 8-10 nuclei arranged in horse shoe shape pattern and at focal areas resorbing dead bone.

Differential count of a peripheral smear showed increase in the lymphocytes up to 56%. Erythrocyte sedimentation rate (ESR) was also elevated to 42mm/hr. The chest radiograph did not reveal any lung pathology. Monteux test was positive and showed 6mm of induration around the inoculation after 24hrs. Tridot test for HIV was done and the patient was non-reactive. A final diagnosis of tuberculous osteomyelitis was made. Hemi maxillectomy had been done and obturator given. 6months follow up has been done and the wound healed uneventful.

Discussion
Tuberculosis is a systemic disease worldwide especially in developing countries like India. Its incidence is increased in HIV patients. Primary tuberculosis affects children and adolescent and usually cervical lymph nodes will be enlarged. Oral tuberculosis are generally caused by direct contact of ulcerated mucosa with infected sputum. The intact epithelium of oral mucosa and presence of salivary enzymes, antibodies are considered as a protective barrier for the entry of tuberculous bacilli. Any breach in the epithelium due to chronic irritation or decreased immunity may help the organisms to colonize in the oral cavity.

Oral tuberculosis lesions account for 0.1-5% of the infections caused by mycobacterium tuberculosis. Clinical presentation of oral
tuberculosis is nonspecific and may present as ulcers or granulomatous or nodular growth. Almost 93% of the oral lesions are ulcers and about half of them affect the tongue. Oral lesions commonly present ulcer of irregular, indurated margins with undermined edges with necrotic bone. Usually they are solitary and painful. These ulcerations last for more than three weeks and increase slowly in size without responding to the topical treatment. It may present as an ulcer, gingivitis, tuberculosis involving periapical tissue, extraction socket, TMJ, maxillary sinus and rarely tuberculous osteomyelitis of mandible and maxilla. Mandibular lesions are common compared to maxilla. Tuberculosis of the jaw presents as a asymptomatic swelling with slow necrosis of the bone and sub periosteal abscess formation resulting in pus discharge through the sinus opening intra orally. Severity of the disease depends on the quantity of bacilli observed.

Osteomyelitis is an inflammatory condition which involves the cortical and cancellous bone and mandible is the most common site involved in the oral maxillofacial region. In few reported cases, the entire mandible is involved by the slow necrosis of the tuberculosis infection and the involved bone is replaced by the granuloma formation. The radiographic presentation may be blurring of trabeculae with irregular radiolucency. The entry for organisms may be through the large open pulp carious lesion, extraction socket, etc. According to Chapotel, incidence of tuberculous osteomyelitis of mandible is more than 60% in younger patients of age below 15 years.

Jorge Telez reported a case of Tuberculous osteomyelitis with neurofibromatosis and presented a table with 8 cases of tuberculous osteomyelitis of mandible in infancy in his article. Few valuable numbers of cases have been reported with tuberculous osteomyelitis of mandible in the literature. But, one case of tuberculous osteomyelitis of maxilla has been reported in 19 years old girl by Ramesh Gupta. Our present case, tuberculous osteomyelitis of maxilla is also rarest of a rare case reported in the literature which affects primarily the maxilla in 39 years old female patient.

Systemic factors such as immune suppression or increase in the virulence of the pathogens and many local factors like chronic inflammation, poor oral hygiene, local trauma, periodontal diseases and surgical lesions play a vital role in the oral tuberculosis. If oral tuberculosis is noticed, it is important to search for the primary site like lungs, lymphnodes. The radiograph may reveal mixed radio opaque to radiolucency with destruction of cortical plate.

Clinically it should be differentiated from other ulcerative lesions like squamous cell carcinoma, traumatic ulcer, syphilitic ulcer, sarcoidosis, Wegener’s granulomatosis, zygomycosis, and leprosy and treated accordingly. Some of the symptoms like fever, lymph node enlargement, cutaneous changes, chronic cough and excessive fatigue may be helpful to differentiate oral tuberculosis from the other entities. Early diagnosis not only reduces the morbidity of the patient, but also helps in protection of dental professionals and community as the patient is a potential source of transmission. In 27-60% of cases, special stains like Zeill-Neelson stain may help in the diagnosis by the demonstration of microorganisms within the tissue as they are scarce in the tissue specimen.

Conclusion
In spite of wide spread of tuberculosis and its high prevalence in developing countries like India, its oral manifestations are rare. Tuberculous osteomyelitis of maxilla is extremely rare. There is no single confirmatory test for the diagnosis of tuberculosis. Hence, it is important to perform number of investigations to confirm or to rule out the disease. Primary tuberculosis of oral cavity is diagnostic challenge to the clinician, because of its nonspecific signs and symptoms. Dentist has a dynamic role in diagnosing the initial stages of tubercular manifestations in the oral cavity by always excluding these lesions in their routine differential diagnosis. Anti-tubercular drug regime should be followed as soon as it is diagnosed to reduce the morbidity of the patient.

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