PRIMARY TUBERCULOUS OSTEOMYELITIS OF THE MANDIBLE- A RARE CASE REPORT

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Abstract:
Tuberculous osteomyelitis of mandible is an extremely rare condition, particularly in comparison to pyogenic infections and neoplastic diseases involving the mandible. Although a rare occurrence, the differential diagnosis of tuberculous osteomyelitis must always keep in dentist's mind when the routine therapy fails to respond. Here, we report an unusual case of primary tubercular osteomyelitis of the mandible where the biopsy of the lesion alone has lead to near final diagnosis of the case and there after antitubercular therapy subsided the swelling.
Key words: Primary Tuberculosis, osteomyelitis, biopsy.

Introduction:

Tuberculosis is a chronic infectious granulomatous disease caused by *Mycobacterium tuberculosis*[1] which is an aerobic, slender, non-motile, non-encapsulated, non-sporing, rod shaped organism ranging from 2 to 5 μm[2]. The World Health Organization (WHO) estimates that worldwide there are approximately 20 million active cases, of them approximately 3 million people die each year from tuberculosis, of which 80 % are in developing countries[3].

Tuberculous oral lesions are relatively rare occurrence. Oral manifestations occur in approximately 3% of cases involving long standing pulmonary and/ or systemic infection[4].Oral clinical presentation may be as ulcers, erythematous patches, and indurated lesions with granular surface, nodules, and fissures or as jaw lesions. The most common sites involved are tongue, gingiva, tooth sockets and jaw involvement may present as osteomyelitis [5].

Two main types of tubercular infections of oral tissues are recognized - Primary and Secondary. Primary lesions develop when tuberculosis bacilli are directly inoculated into the oral tissues of a person who has not acquired immunity to the disease and in fact, any area that is vulnerable to direct inoculation of bacilli from exogenous source can be a potential site. These frequently involve gingiva, tooth extraction sockets and buccal folds. Secondary infection of oral tissues can result from either haematogenous or lymphatic spread or from autoinoculation by infected sputum and direct extensions from neighbouring structures. Intraoral sites frequently involved include the tongue, palate, lips, alveolar mucosa and jaw bones[2].

With myriad presentations and sometimes lack of specific systemic symptoms, oral tubercular lesions may present as puzzle for us and may escape our eyes. Hence, we document a case of primary tuberculous osteomyelitis of mandible in an old male individual who was initially suspected for dental abscess with nonspecific chronic osteomyelitis and later proved as primary tubercular osteomyelitis.

Case report

A 54 year old male patient presented with a gradually increasing swelling on the right mandibular region of face since 2 months. The patient reported that 3 months back he had visited a dentist for severe pain associated with 46 and extraction of 46 and 27 was done, after which swelling started. He had received multiple courses of oral antibiotics after which he reported to our institute. Extra oral incision and drainage along with multiple courses of antibiotics were started; despite this the swelling didn't subside. The swelling which was initially small gradually increased in size with extraoral sinus formation and appearance of second swelling above it (Figure 1 &2).

Physical examination revealed that he was well built, nourished and afebrile. On local extraoral examination there was a well defined ovoid swelling of approximately 3x3 cm on the right submandibular region extending

from inferior border of right side of mandible to few cms short of angle of the mandible. Surface appeared erythematous, smooth, shiny, sinus opening with serosanguinous discharge. On palpation it was tender, soft to firm inconsistency, nonfluctuant, surface raise of temperature and was attached to the underlying structures. Lymph nodes were not palpable. The second swelling appeared 2cms above it which was 1x1 cm, tender, erythematous, smooth, firm. On intra oral examination there was no apparent change from normal except for slight expansion of buccal cortical plate in vestibule, below a well healed extraction socket 46 (Figure-3). There was generalized attrition and gingival recession with poor oral hygiene.

A panoramic radiograph revealed an ill-defined radiolucency extending from 45 to 47 postero-inferiorly (Figure 4) and was in contrast to the initial radiograph provided by the patient which showed a well defined radiolucent area surrounding the roots of 46 for which he was treated (Figure 5). A provisional diagnosis of chronic nonspecific osteomyelitis was given and since it was refractory to medication, actinomycosis and tuberculosis were thought of as differential diagnosis.

On further evaluation he revealed, he had dry cough since 6 months, didn't notice evening rise of temperature, weight loss and was not aware of possible contact with tuberculosis persons.

Routine laboratory tests and investigations for tuberculosis were simultaneously carried out. All haematological values were within the normal limits, ELISA for HIV was negative. A tuberculin (Montoux) test was negative, Chest radiograph (PA view)(Figure-6) did not reveal any abnormal findings, Test for C-reactive protein was also negative, sputum examination for Acid fast bacilli (AFB) did not reveal any bacilli and culture after 48 hrs of incubation was negative. Cytological smear taken from the sinus drainage was stained with Ziehl-Neelsen, PAS, but revealed only polymorphonuclear leucocytes, few lymphocytes in necrotic background and no actinomycotic colonies or bacilli were detected. FNAC showed no granulomatous lesion.

Since the tests were inconclusive, biopsy from extraction socket and curettage of the lesion was performed and sent for histopathological examination (Figure-7). It showed granulomas of varying size and shape consisting of central Langhan's giant cell, epitheloid cells, surrounded by lymphocytes and few plasma cells (Figure-8). Few granulomas showed extensive caseating necrosis in the centre (Figure 9). Areas of necrosis with polymorphonuclear neutrophills were also appreciated. The above features were consistent with tuberculous granuloma, a chronic granulomatous lesion. For confirmative diagnosis sections were stained with Zeihl-Neelsen, which did not reveal any bacilli and specimen was sent for culture.

Standard antitubercular therapy for bone tuberculosis was started. At 2 weeks follow up some reduction in size of swelling was noticed (Figure 10).

Discussion

Tuberculosis (TB) remains the leading cause of death worldwide from a single infectious organism[1]. It is very common in India and South-East Asia, where the prevalence rate is about four in every 1,000 people.15% of tuberculous population of the world resides in India [6]. Primary oral tuberculosis is rare, as an intact oral mucosa, cleansing action of saliva, salivary enzymes, tissue antibodies and oral saprophytes act as barriers to infection. Any breach in these defense mechanisms, such as abrasions, tears, chronic inflammation, poor oral hygiene, tooth eruption, extraction sockets, periodontal disease, and carious teeth with pulp exposure may lead to infection by, tubercle bacilli[4, 5] .Poor socio-economic conditions with inadequate nutrition and lack of hygiene are predisposing factors to infection [5].

Bone TB is a relatively uncommon form of extrapulmonary tuberculosis seen in approximately 1% of children with TB [7]. It is more frequently seen in children as compared to adults because of highly vascularized bone in infants and children [8]. Tuberculous osteomyelitis is quite rare and constitutes less than 2% of skeletal TB. Jaw involvement is even rarer and affects older individual [3,9,10] and also children [3, 5]

The involvement of the mandible by TB infection is extremely rare as it contains less cancellous bone. But the mandibular involvement is more frequent than maxilla[11] and the alveolar and angle regions have greater affinity. The infection may extend to the mandible by:

1) Direct transfer from infected sputum or infected raw milk of cow through an open pulp in carious tooth, an extraction wound or gingival margin or perforation of an erupting tooth
2) Regional extension of soft tissue lesion to involve the underlying bone
3) Haematogenous route [11]

Chapotel [12] described four clinical forms of tuberculosis of the mandible.

1. *The superficial or alveolar form* in which the alveolar process is involved either by direct extension of the tuberculous gingival tissues or by way of a deep carious tooth. The course is usually chronic, and necrosis of bone is progressive, with the formation of abscesses and fistulae.
2. *The deep or central form*, in which the lesion involves the angle of the mandible. It is found, according to Chapotel, almost exclusively in children during the period of eruption of the molar teeth.

3. *The diffuse form*, characterized by progressive extensive necrosis of mandible, which at times involves the tempromandibular articulation following a period of swelling and suppuration. Painless pathological fracture may occur. Severe general symptoms, accompanying a wide spread of tuberculosis affecting the liver, the lungs, the kidneys, and the meninges, are characteristic of the fatal aspect of this form.
4. *The acute osteomyelitis form*, in which, as the name implies, the sudden onset, the acute local and general manifestations, and the rapid course simulate those of an acute osteomyelitis of the mandible. This form is, however, very rarely obsereved[12].
5.

There have been cases of primary TB of the mandible reported in adults and they did not find any primary focus in lung [10]. Meng [13] stated that jaw infection is practically always associated with pulmonary infection and 43% of his cases had TB of other bones. But in our case there was no history of pulmonary or osseous involvement. Hence the spread of infection might have been through an extraction socket of 46, which could have made an occult tubercular focus in the mandible. Moreover, the patient was of lower socio-economic conditions with poor oral hygiene which might have been a predisposing factor.

Tuberculosis of the jaw causes slow necrosis of the bone and may involve the entire mandible[11].There is no characteristic radiographic appearance of TB of the jaws, or alveolar bone and most lesions are indistinguishable from those caused by pyogenic organisms [14]. The destruction of the bone in radiographs appears as blurring of trabecular details with irregular areas of radiolucency. There is an erosion of the cortex with little tendency to repair. Gradually the bone is replaced by soft tuberculous granulation tissue. Caseation appears at places followed by softening and liquefaction. A subperiosteal abscess forms presenting as a painless, soft swelling. This cold abscess may burst either intra or extraorally forming single or multiple sinuses. Pathological fracture of mandible and sequestration may also occur [11].

Meng [13] stated that, district where TB is prevalent, if young patient presents a painful swelling of the jaw, following an insidious onset with or without preceding attacks of toothache, discharging sinus, roentgenogram shows a varied extent of necrosis of bone without much evidence of involucrum, one must look out for a tuberculous lesions of the mandible.

India being a country with high prevalence of TB in population, our case which presented a painful swelling with preceding toothache, discharging sinus, multiple swellings, necrosis of bone with no involucrum on radiographic picture, refractory to oral antibiotics led us to tuberculous osteomyelitis as one of the differential diagnosis.

Traditionally, the diagnosis of tuberculosis has been made on the basis of clinical findings and radiographs and confirmed by sputum or tissue smears that show AFB bacilli. These methods remain the gold standard for diagnosis, but the development of DNA probes, polymerase chain reaction assays, and liquid media now allow, more sensitive and rapid diagnosis [3]. However our country being a developing country, the facilities is scarce and expertise required are lacking. FNAC has been a useful adjunct in the diagnosis of TB and is the first choice of investigation in India [14, 15].

Absence of AFB in smears, FNAC, showing an otherwise characteristic cytological picture should not weigh against the diagnosis of TB [14, 15]. The smears and culture for AFB, from the oral lesions and the sputum, were negative in two cases of primary tuberculosis of the oral cavity reported by Dimitrakopoulos I et al 1991 [9]. They quoted various reasons cited by different authors for the difficulty in the microbiologic detection of the tubercle bacilli. This may be due to

1) High immunity of the patient resulting in the destruction of the bacilli
2) Their enclosure by local tissue reaction and the very small numbers of tubercle bacilli in the oral lesions
3) Previous long time treatment with antibiotics

Conclusion:

The case reported in this paper emphasizes the importance of histopathological diagnosis in any long standing swellings refractory to routine treatment of which tuberculosis is one. If detected early, the bony changes can be reversed without much destruction and therefore there should be prompt initiation of an effective regimen without wait and watch attitude even though there was lack of confirmation, as in our case and which regressed after therapy.

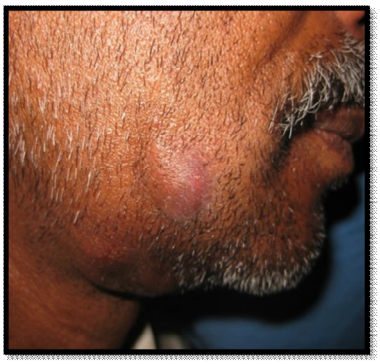

Figure 1: Extraoral swelling

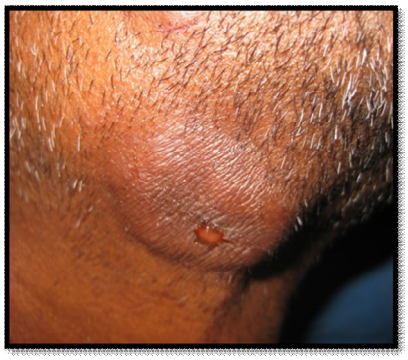

Figure 2: Extraoral sinus formation

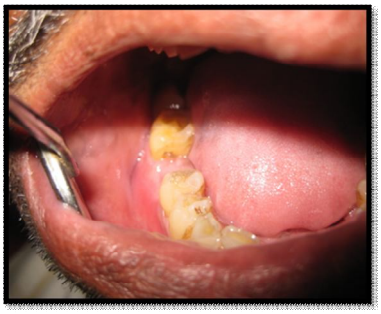

Figure 3: Intraorally well healed socket

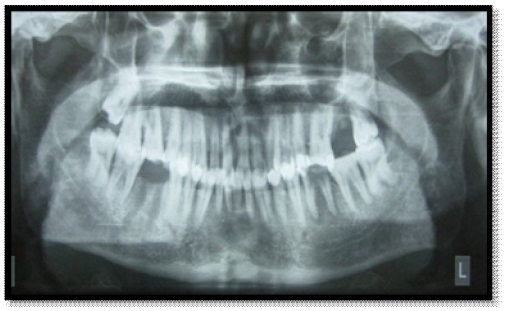

Figure 4: An ill-defined radiolucency extending from 45 to 47 postero-inferiorly

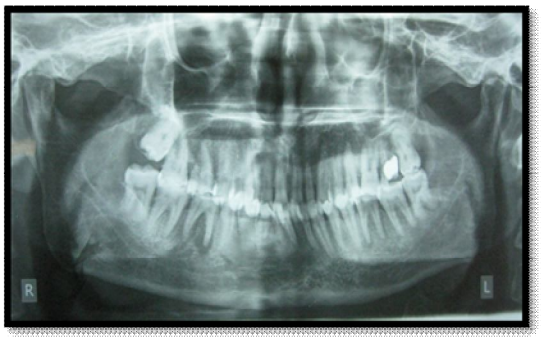

Figure 5: Well defined radiolucent area surrounding the roots of 46 for which he was treated

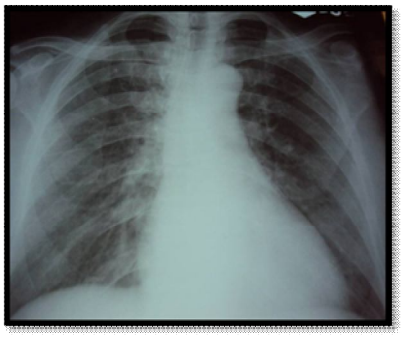

Figure 6: P.A View of chest

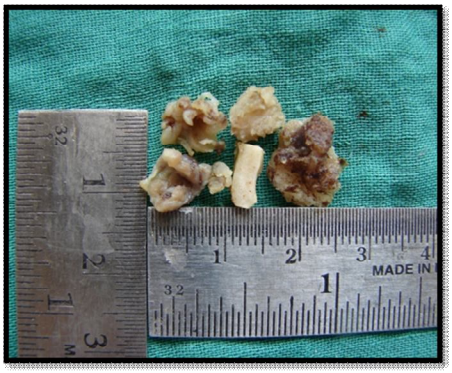

Figure 7: Gross specimen

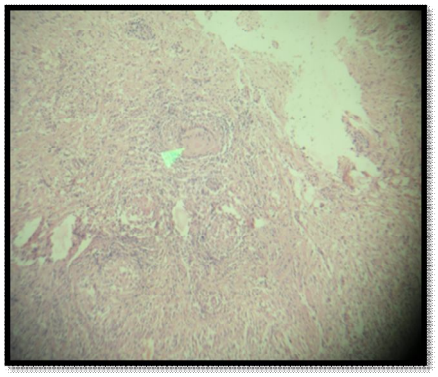

Figure 8: Central Langhan's giant cell, epitheloid cells, lymphocytes and few plasma cells (4x)

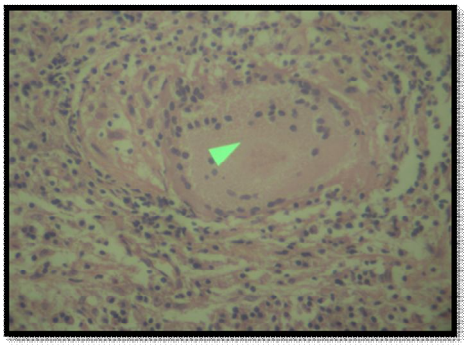

Figure 9: Caseating necrosis in the centre (40x)

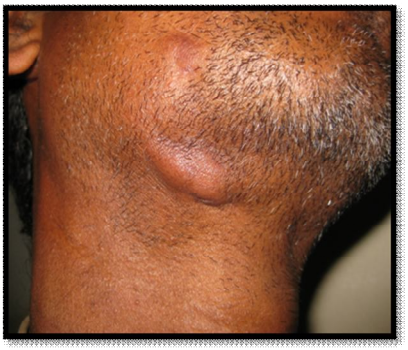

Figure 10: 2 weeks follow up shows some reduction in size of swelling was noticed

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