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Case report

Diagnostic dilemma in temporomandibular joint tuberculosis: A case report

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ABSTRACT: Extra-pulmonary tuberculosis (TB) of the temporomandibular joint (TMJ) and orofacial region is a diagnostic challenge and a high index of suspicion is required for a correct diagnosis. A patient was referred to us with a provisional diagnosis of a parotid fistula in the left submandibular region. Further evaluation revealed this to be TB of the left TMJ with involvement of the adjoining muscle. We report this case of TB involving the TMJ because of its rarity and the associated diagnostic dilemma.

Key Words: Extra-pulmonary tuberculosis, osteomyelitis condyle, fine needle aspiration cytology, cervical lymphadenopathy, acid-fast bacilli.

INTRODUCTION:

Tuberculous (TB) lesions of the orofacial region resulting from primary inoculation are unusual. Most cases are secondary to TB in other parts of the body, usually the lungs [1]. Primary infection may occur in other organs, including bones and joints, but are mostly seen in the major joints [2]. TB of temporomandibular joint (TMJ) has been reported occasionally [2, 3].

Extra-pulmonary tuberculosis is defined as TB of a region other than the lungs, such as the pleura, lymph nodes, skin, and joint spaces [4]. extra-pulmonary TB comprises 15~20% of all cases of TB among immunocompetent adults, and more than 50% of the cases in human immunodeficiency virus-positive individuals [5]. Reports from different parts of the world indicate an increase in the proportion of extra-pulmonary TB among all TB cases [6]. TB of the jawbones may be secondary or primary and occurs as a result of deep extension of a gingival lesion, from an infected post-extraction socket, or through haematogenous spread of infection [7]. The cancellous component of the mandibular condyle makes it prone to TB involvement.

CASE REPORT:

A 45-year-old man, who was non-diabetic, non-hypertensive, a non-smoker, non-alcoholic, and had a low socioeconomic status, presented with pain, swelling, and discharge from a non-healing incision in the left submandibular region. He was unable to open his mouth wide (trismus). He had previously undergone incision and drainage of a parotid abscess and prolonged parenteral antibiotic therapy. There was a history of fever, anorexia, and moderate weight loss. There was no history of trauma to the affected region. The patient had an ambiguous history of anti-tubercular treatment (ATT) in childhood for TB of the lung. There was a watery discharge from the wound that was not foul smelling (Fig. 1).

Figure 1: There is non healing discharging wound in the left submandibular region.

During the examination, the trismus yielded to forceful mechanical opening, but this was very painful. The patient was started on physiotherapy for mouth opening and an orthopantomogram (OPG) was obtained.

High-resolution computed tomography (HRCT) of the TMJ, face, and neck revealed irregularity of the cortical outline with patchy lysis and demineralisation of the mandibular condyle and neck, with deformation of the glenoid fossa and condylar head. There was a loss of joint space with appreciable soft tissue changes in the adjacent muscles (Fig. 2a, 2b).

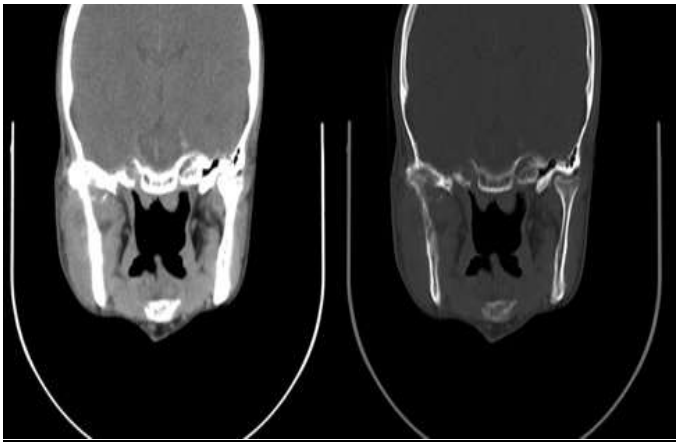

Figure 2: (A and B) High-resolution computed tomography (HRCT) of the temporomandibular joint (TMJ), face, and neck revealed irregularity of the cortical outline with patchy lysis and demineralization of the mandibular condyle and neck, with deformation of the glenoid fossa and condylar head. There was a loss of joint space with appreciable soft tissue changes in the adjacent muscles.

A routine anteroposterior chest X-ray suggested pulmonary Koch's syndrome. The laboratory findings were within normal limits, with an elevated erythrocyte sedimentation rate (Westergren method) of 24 mm at the 1st hour, elevated alkaline phosphatase at 186 U/L, and a positive Monteux test with a 20 × 20 mm induration. The collagen profile, including rheumatoid (RA) factor, anti-nuclear antibodies, and anti-cyclic citrullinated peptide, was negative. The serum angiotensin-converting enzyme (ACE) was within normal limits. The wound discharge was sent for pyogenic culture and sensitivity tests. The acid-fast bacilli (AFB) smear examination and cultures were negative. The subject tested negative for retroviruses. In the absence of another probable diagnosis, such as fungal infection, actinomycosis, malignancy, or sarcoidosis, the patient was provisionally diagnosed with smear-negative pulmonary TB and TB of the TMJ joint.

The ATT was started with daily rifampicin 600 mg, isoniazid 300 mg, ethambutol 1,000 mg, and pyrazinamide 1,500 mg with pyridoxine 20 mg. At follow-up, his symptoms had improved, with resolution of the submandibular wound and swelling. The mouth opening improved, but with leftward deviation on opening. In the course of clinical resolution, a small abscess appeared on the skin surface overlying the masseter muscle; the original sinus tract healed, but a new sinus tract appeared superiorly over the subcondylar region. Follow-up OPG after 4 months of ATT revealed a pathological fracture of the left condylar neck (Fig. 3).

Figure 3: Follow-up orthopantomogram (OPG) shows separation of the left condylar head and neck along with deformation of the left TMJ.

The sequestered bone, formed due to pathological fracture of the left condyle, might have caused the persistent sinus to shift superiorly. Any surgical intervention of the left TMJ was planned to be delayed until 6 months after the ATT had been completed.

DISCUSSION:

Primary extra-pulmonary TB represents 17% of all reported cases of *Mycobacterium tuberculosis* infection in Canada, and as much as 30% of cases in the United States [8]. Of the reported cases of extra-pulmonary TB, 6~15% were primary infection of bone or joints [9]. There are few reliable epidemiological studies on the relative contributions of pulmonary and extra-pulmonary disease to the total number of TB cases in India [5], although reports of extra-pulmonary forms have increased recently.

Most reported cases of TB of the jaws are primary and can appear as lesions on the gingiva, palate, lips, tongue, buccal mucosa, frenulum, and jawbones; rarely, secondary oral manifestations are associated with pulmonary infection [2]. Possible modes of dissemination are direct transfer, regional extension of soft tissue lesions, haematogenous and lymphatic routes, or a combination of these modes. Direct transfer can occur through open pulp, extraction wounds, the gingival margin, or perforation of an erupting tooth.

The most frequent manifestation of head and neck TB (i.e. in 95% of cases) is cervical lymphadenitis [10]. Jaw involvement in TB is rare and usually affects older individuals [11].

Our case of TB of the condyle deserves special attention because of the extra-pulmonary TB and nonspecific findings, which were similar to those of arthritis, osteomyelitis, chronic joint disease, or malignancy of the TMJ. The most common symptom is preauricular swelling (unresponsive to antibiotics) associated with trismus [3]. Involvement of the mandible by TB is rare, as it contain less cancellous bone. It has been suggested that TB of the TMJ is most often misdiagnosed as arthritis [6].

In the reported cases of TMJ TB, including our patient, pain, trismus, and swelling were the chief clinical features [2, 9,12-14]. TMJ TB should be considered in the differential diagnosis of patients presenting with pain and stiffness of the joint [2,15].

The gold standard for the diagnosis of osseous TB is culturing *M. tuberculosis* from bone tissue. The presence of TB granulomas and AFB is an important diagnostic clue. The paucibacillary nature of the disease means that it is sometimes difficult to find AFB in smears [16]. Fine needle aspiration cytology (FNAC) is a minimally invasive tool that has an established role in the diagnosis of extra-pulmonary TB, as well as oral lesions. We considered ultrasound-guided FNAC of the lesion, but negative findings of TB might have been due to the procedure itself. Even when adequate tissue is procured, the pathology may suggest "granulomatous infection", which encompasses a wide differential diagnosis, rather than "definitive tuberculosis". It should be remembered that while a positive non-conventional test may "rule in" a diagnosis, a negative test cannot "rule out" a diagnosis of TB [5]. Type I, II, and III epithelioid granulomas are 7.4, 35.6, and 54.2%

positive for AFB, respectively [17]. The Monteux test is positive in more than 90% of cases of osteoarticular TB [18].

TB of the joint must also be differentiated from RA. TB is usually monoarticular in nature, whereas RA is polyarticular. The treatment of TB bone infection involves a complete course of anti-tuberculosis drugs. Surgical excision and decortication are reserved for the most refractory cases if intense pharmacotherapy fails [9]. An early diagnosis and treatment of osteoarticular TB can achieve 90~95% cure with nearly normal function [19]. Delayed or missed diagnosis and treatment can result in osteoarthritic changes and severe joint destruction [2].

The diagnosis of TB of the maxillofacial region is made according to the combination of strong clinical suspicion, especially in endemic areas, clinical evidence of the disease, and the results of cytological/histopathological examinations. An early diagnosis is of critical importance to ensure less destruction and full functional rehabilitation of the TMJ.

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