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Tuberculosis of Temporomandibular Joint Presenting as Swelling in the Preauricular Region

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Extrapulmonary tuberculosis (EPTB) is defined as tuberculosis (TB) of organs other than the lungs, such as the pleura, lymph nodes, abdomen, genitourinary tract, skin, joints, bones, and meninges.[1] EPTB constitutes 15% to 20% of all cases of TB among immunocompetent adults, and it accounts for more than 50% of the cases in human immunodeficiency virus (HIV)–positive individuals.[2] Reports from different parts of the world indicate an increasing trend in the proportion of EPTB among all TB cases.[3] The HIV pandemic,[4] improved case recruitment because of expanding services,[5] reduction in infectious TB,[3] and immigration resulting in changing demographics[6] have been cited as causes for the increasing proportion of EPTB. Therefore, it is imperative that clinicians recognize the clinical signs and symptoms of EPTB, make a prompt and accurate diagnosis, and properly treat the disease.

Osteoarticular TB is commonly seen in the spine and the large weight-bearing joints, such as the hip, knee, and ankle. Joint involvement occurs by hematogenous spread from a pulmonary, visceral, or lymph node focus leading to bacterial colonization of vascular cancellous bone, especially the epiphysis and metaphysis of long bones.[7] Infection begins in the subchondral region and spreads to involve the cartilage, synovium, and joint space.[8]

Although head-and-neck TB comprises nearly 10% of all extrapulmonary manifestations of the disease,[9] TB of the oral cavity and the maxillofacial region is rare.[10] Tuberculous involvement of the temporomandibular joint (TMJ) is uncommon, but has been reported occasionally.[9,11] The cancellous component of the mandibular condyle makes it prone to tubercular involvement. Rare lesions such as TMJ TB may pose a diagnostic challenge, but need to be considered in light of the increasing incidence of EPTB.

Case Report

A 45-year-old man complained of a swelling on the right side of the face for 1 month (Fig 1). On careful inquiry, the patient reported that he had had difficulty chewing for the last 3 months and mild swelling developed approximately 2 months previously, with rapid growth in the last month. The patient was febrile over the past 3 weeks. He consulted a local dentist, who prescribed antibiotics and analgesics, which brought him no relief. Otherwise, the patient's medical history and intraoral examination were noncontributory. On clinical examination, he had a preauricular scar after trauma around 15 years previously. The swelling was tender and firm, and it extended from the preauricular region to the zygomatic arch. The temperature of the overlying skin was normal. His mouth opening was reduced to less than the width of 2 fingers.

The differential diagnosis included infection and neoplasm arising from the TMJ or the parotid gland. Suppurative arthritis, osteomyelitis, osteoblastoma, osteosarcoma, and malignant lesions of the parotid gland were also considered in the differential diagnosis.

To aid in diagnosis, routine blood investigations, a panoramic radiograph, fine-needle aspiration cytology (FNAC) and magnetic resonance imaging (MRI) were performed. Blood test results were normal except for the erythrocyte sedimentation rate, which was 72 mm for the first hour (normal value, 14 mm/h). The patient was seronegative for HIV and hepatitis B and C. The panoramic radiograph showed erosion of the right condyle and widening of the glenoid fossa, whereas the left-side TMJ was normal (Fig 2). Results of aspiration cytology showed scattered granulomas composed of epithelioid cells and some lymphocytes (Fig 3). Necrotic debris was also identified in some areas. Ziehl-Neelsen stain showed occasional acid-fast bacilli (AFB) (Fig 4). MRI with contrast showed peripheral enhancement of the right TMJ and surrounding muscles resulting from caseating granuloma formation and diffuse inflammation (Fig 5). When combined, these findings suggested tubercular infection.

The patient was referred to a physician for further consultation and clinical and diagnostic tests. Although the

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FIGURE 1. Photograph showing preauricular swelling on right side of patient.

Ranganathan et al. Tuberculosis of Temporomandibular Joint. J Oral Maxillofac Surg 2012.

Mantoux test was positive, normal chest and spine radiographs, computed tomography scan of the abdomen, and the sputum culture were all negative, thereby ruling out pulmonary or any other tubercular focus in the body. Hence, a diagnosis of primary TB of the condyle was made.

After the definitive diagnosis was made, the patient was prescribed a 3-month antitubercular regimen consisting of oral rifampicin, 600 mg/d; isoniazid, 300 mg/d and pyrazinamide, 1750 mg/d; and steptomycin injection,1000mg/d. Streptomycin was discontinued, and the rest of the drugs were continued for 6 more months. After 9 months of the antitubercular drug therapy, the condition fully resolved. The 1-year follow-up examination showed no recurrence of the disease and showed normal mouth opening and functional movements.

FIGURE 2. Panoramic radiograph showing erosion of condyle of right TMJ and widening of glenoid fossa.

Ranganathan et al. Tuberculosis of Temporomandibular Joint. J Oral Maxillofac Surg 2012.

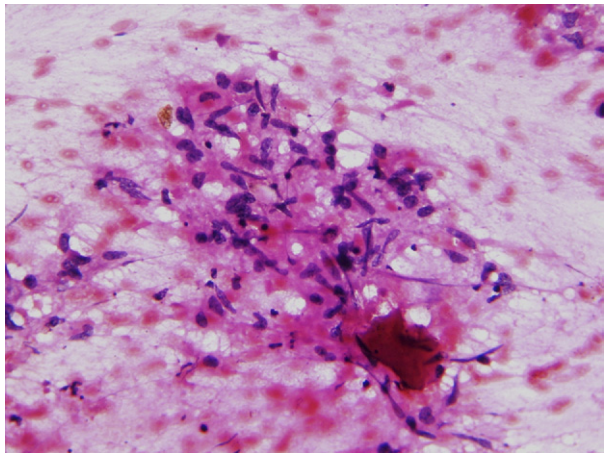

FIGURE 3. Photomicrograph of aspiration smear showing epithelioid cell granuloma (hematoxylin-eosin stain, original magnification ×400).

Ranganathan et al. Tuberculosis of Temporomandibular Joint. J Oral Maxillofac Surg 2012.

Discussion

Osteoarticular TB is an extrapulmonary form of TB that can be primary or due to a pulmonary focus. It usually involves the spine and long bones. Because of the absence of classic symptoms associated with pulmonary disease, such as fever, cough, weight loss, anorexia, and night sweats, diagnosing extrapulmonary TB has often been a clinical challenge. Osteoarticular TB is usually monoarthridal and involves the cancellous portions of the bone and, hence, frequent involvement of the metaphysis and epiphysis of the long bones.[12] Hematogenous dissemination and trauma are usually considered the major etiologic factors for osteoarticular TB.

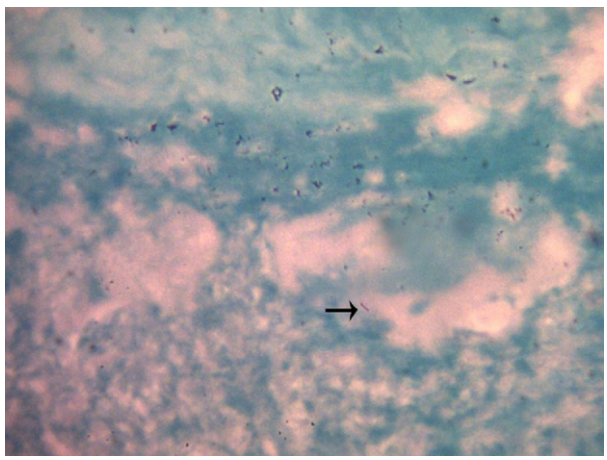

FIGURE 4. Photomicrograph showing AFB (*arrow*) (Ziehl-Neelsen stain, original magnification ×1,000).

Ranganathan et al. Tuberculosis of Temporomandibular Joint. J Oral Maxillofac Surg 2012.

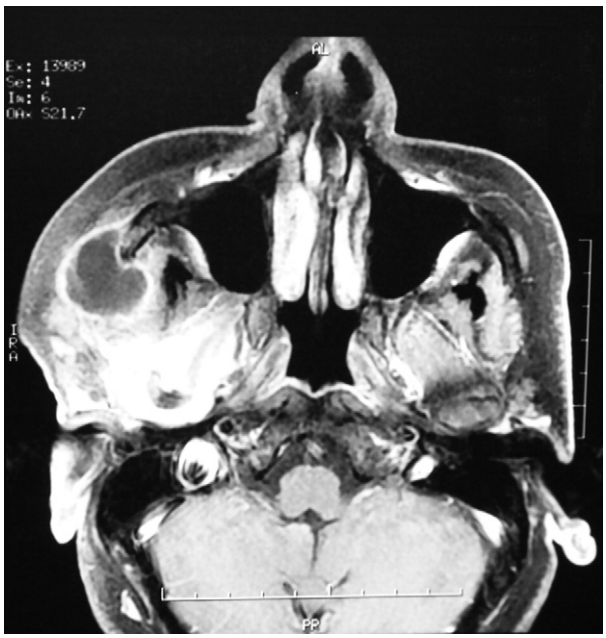

FIGURE 5. MRI study showing peripherally enhancing collection of right TMJ and surrounding muscles.

Ranganathan et al. Tuberculosis of Temporomandibular Joint. J Oral Maxillofac Surg 2012.

Oral TB is relatively uncommon, with frequent involvement of the buccal mucosa, tongue, gingiva, and mandible. The intact mucosa acts as a natural barrier to the mycobacterial invasion because of its epithelial thickness, tissue antibodies, oral saprophytes, and salivary enzymes, as well as cleansing action of the saliva. Any break in continuity of the mucosa due to trauma or any other pathology may result in mycobacterium invasion of deeper tissue.[13] Despite very few cases of oral TB in patients with positive sputum samples, local injury is blamed for the progression of the tubercular infection from the oral cavity to the jaw bones.[14] As noted, very few cases of tubercular involvement of the TMJ have been reported in the literature[11,15-18]; it has been suggested that the condition is often misdiagnosed as arthritis.[3]

In all the reported cases of TMJ TB, including our patient, pain, trismus, and swelling are the clinical features.[11,15-18] Thus, TMJ TB should be considered in the differential diagnosis of patients presenting with pain and stiffness of the joint.[11,19] Most of the time, swelling and pain are misdiagnosed as odontogenic infection and the patients are prescribed antibiotics.

Ruggiero et al[17] stressed that benign and malignant neoplasms of the joint and infective processes such as acute suppurative arthritis, osteomyelitis, and chronic tuberculous arthritis have to be considered in such cases. Because the TMJ is closely associated with the ear and major salivary glands, exploration and biopsy are necessary to rule out pathology associated with these structures and to help establish a definitive diagnosis.

Most nonpulmonary forms of primary TB emanate from hematogenous dissemination.[9] The origin of the TMJ infection in our case remains unclear. As previously noted, the patient had a history of trauma to the side of the face, so trauma cannot be ruled out as the etiologic factor.

Increasing incidence of multidrug-resistant tuberculosis (MDR-TB) is also posing a challenge to clinicians. It is estimated that approximately 440,000 persons had MDR-TB worldwide in 2008 and that one-third of them died of the disease. Asia accounts for the majority of cases; it was estimated that almost 50% of MDR-TB cases worldwide occur in China and India.[20] It is reasonable to suggest that the emergence of MDR-TB is because of medical error. MDR-TB may result from prescribing an unreliable regimen or unreliable drugs or failing to ensure (by directly observing treatment and education of the patient and his or her family) that the patient takes the drugs as prescribed and for the full prescribed period. MDR-TB results from a failure of effective implementation of national TB programs. Treatment of patients with MDR-TB (especially those with resistance to rifampin and isoniazid) may have to involve "second-line" reserve drugs, that is, drugs other than the "standard" essential antituberculosis drugs (ie, rifampin, isoniazid, streptomycin, ethambutol, pyrazinamide, and thioacetazone).[21]

The gold standard for the diagnosis of osseous TB is culture of *Mycobacterium tuberculosis* from bone tissue. Showing TB granulomas and AFB is important for the diagnosis of TB. Because osteoarticular TB is paucibacillary in nature, it sometimes becomes difficult to show AFB in the smears.[22]

FNAC is a noninvasive tool and has an established role in the diagnosis of extrapulmonary TB, as well as in oral lesions. We considered FNAC a suitable means of diagnosis.[23] Cytology smears should show the epithelioid granuloma with or without necrotic material. Epithelioid granulomas are usually classified into 3 types. Type I is epithelioid granuloma without necrosis, type II is epithelioid granuloma with necrosis, and type III is necrosis without epithelioid granuloma. Sometimes, there is intense neutrophilic infiltration in the necrotic material, which indicates an acute suppurative lesion. The rates of AFB positivity in type I, type II, and type III granulomas are 7.4%, 35.6%, and 54.2%, respectively.[24]

The Mantoux test is positive in more than 90% cases of osteoarticular TB.[25] However, a positive test may also indicate a hypersensitivity reaction to tuberculin proteins or a previous exposure rather, than active tubercular infection.

Radiographic findings show erosion of the condyle and glenoid fossa. The combined presence of swelling, trismus, functional limitations, and radiographic evidence of erosion of the TMJ would suggest that clinicians consider a differential diagnosis of infection and neoplasm.[17]

Radiologically, TB of the joint must also be differentiated from rheumatoid arthritis (RA). TB is usually monoarticular in nature, whereas RA is polyarticular. MRI features of osteoarticular TB include increased synovial membrane thickness, bone erosions, bone marrow edema, and extra-articular cystic collections. Synovial membrane thickness in TB is less than that in RA. However, the size of bone erosions and rim enhancements around erosions is greater in osteoarticular TB than in RA. MRI findings also show bone marrow edema and extra-articular cystic collections in osteoarticular TB.[26]

Thus, diagnosis of TB of the maxillofacial region results from a combination of strong clinical suspicion (especially in epidemic areas), clinical evidence of the disease, and the results of cytologic/histopathologic examinations. When involvement of bone is suspected, imaging is important.

The treatment of tuberculous bone infection involves a long course of antituberculous drugs. Surgical excision and decortications are reserved for the most refractory cases, when intense pharmacotherapy has failed.[17] Early diagnosis and treatment of osteoarticular TB can achieve 90% to 95% cure with nearly normal function.[27] Missed or delayed diagnosis can result in osteoarthritic changes and severe joint destruction.[11] Awareness of the clinical features of this disease, combined with adequate history taking, clinical examination, and investigations in all clinical care settings, is of critical importance to ensure lesser destruction and fully functional rehabilitation of the TMJ.

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