CASE REPORT

Tuberculous osteomyelitis of mandibular condyle: a diagnostic dilemma

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The incidence of tuberculosis (TB) is increasing worldwide and so are its consequences. Its oral manifestations are infrequent, occurring in approximately 3% of all cases. Although the primary lesion occurs as a pulmonary infection, the extrapulmonary infections have also shown an increase over the past few years. These infections generally involve the head and neck through haematogenous or lymphatic routes. The clinical presentation may be as an ulcer, granuloma, orofacial TB, TB of the salivary glands or tuberculous lymphadenitis. Rarely, secondary oral manifestations associated with pulmonary infection are seen, which can appear as lesions on the gingiva, palate, lips, tongue, buccal mucosa, frenulum and in the jaw bones. Owing to the rarity of orofacial TB, it seldom arouses clinical suspicion, especially when a positive history of a systemic infection or therapy is denied. Tuberculous involvement of the mandibular condyle is even rarer, and only two such cases are reported so far, both in English-language literature. Further, the diagnosis of such a case is extremely difficult as there are no specific signs pathognomonic of infection. The only manifestation may be a localized painful swelling of the jaw. The presented case is of osteomyelitis of the mandibular condyle in a 20-year-old male patient in whom TB was later suspected. In this case report the role of diagnostic techniques is emphasized as the osteomyelitis of the condyle has the risk of being easily missed owing to its atypical signs and symptoms and atypical radiographic appearance.

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Case report

A 20-year-old male patient presented with a gradually progressive swelling at the side of his face in front of the right ear for about 15 days (Figure 1). 2 months beforehand, there was a sudden onset of dull pain at the right temporomandibular joint (TMJ) region which aggravated during mouth opening; it was associated with reduced mouth opening. The patient's medical history was non-contributory. There was no history of any trauma to the TMJ region. He was moderately built and nourished, and afebrile at the time of examination. He otherwise reported weakness and malaise over the past month along with some weight loss. General physical and systemic examinations were unremarkable. All the vital signs were in normal range. He was a gutka chewer

(a preparation of tobacco mixed with areca nut) and had chewed it 5–6 times a day for 3 years.

Extraorally, a unilateral solitary swelling was seen at the right TMJ region measuring 1.5 × 1.5 cm in size. It was oval with normal overlying skin. On palpation, the swelling was firm, tender and non-fluctuant. There was a localized rise in temperature. Tenderness was elicited in the temporalis muscle of the same side with no obvious lymphadenopathy of the head and neck region. Intraorally, the buccal mucosa of the left side was opaque and blanched. There were no other significant findings. The occlusion was not affected. On palpation, thick fibrous bands were felt in the left buccal mucosa. This gave a clinical impression of oral submucous fibrosis which could be a contributory factor for the patient's reduced mouth opening apart from the swelling.

Suspecting a middle ear infection, the patient was referred to an otolaryngologist for otoscopy. The findings were not significant.

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Figure 1 Patient with swelling in the right pre-auricular area

Radiographic findings

On the digital panoramic radiograph (shot by Orthophos XG 5 DS Ceph; Sirona, Bensheim, Germany) there was an ill-defined area of radiolucency seen in the right condyle (Figure 2).

The resorption of the condyle was suggestive of osteomyelitis.

Ultrasonography (USG) of the right parotid region was then advised. USG was done on Logiq 500 Pro (GE Medical Systems, Milwaukee, WI) with a high frequency linear array transducer (9.6 MHz frequency).

Ultrasonographic findings

The parotid gland appeared normal. The condyle and neck of the mandible of the right side showed irregularity with erosions suggestive of abscess formation (Figure 3). One lymph node anterior to the tragus on right side was enlarged and hypoechoic. Thus, osteomyelitis of the condyle and ramus of the mandible of the right side with abscess formation was suspected.

A CT of the head was taken after injecting 60 ml of non-ionic intravenous contrast by single slice spiral CT scanner (Somatom Emotion, Siemens, Munich, Germany) at 80 mAs, 130 Kv and 5 mm slice width. A reconstruction of the images was done as required.

CT findings

In the bone window there was evidence of erosion with comminuted destruction of the right mandibular condyle. In the soft-tissue window with contrast there was abscess formation with peripheral enhancement surrounding the right condyle extending into the right masseter and pterygoid muscles (Figure 4). There were a few minimally enlarged right cervical lymph nodes along the jugular chain. The findings correlated to the osteomyelitis of the right mandibular condyle with a surrounding abscess and cervical lymphadenopathy.

Diagnosis

Chronic suppurative osteomyelitis of the condyle was suspected but there was no significant intraoral finding

Figure 2 Panoramic radiograph showing a radiolucent lesion involving the right condylar process of the mandible

Figure 3 Ultrasonography showing abscess surrounding the right condyle of mandible with erosions

related to it. A haematological examination revealed a mildly increased erythrocytic sedimentation rate (ESR: 26 mm h^{-1}) and raised polymorphonuclear leucocytes suggestive of infection. As there was no significant intraoral finding and no history of trauma or operative procedure performed, a systemic source of infection was suspected. Ultrasound-guided fine needle aspiration was done from the dependent part in between the centre and the lower border of the swelling with the help of a syringe. Blood-tinged pus was obtained on aspiration. This was sent for cytopathological

Figure 4 Coronal CT with contrast in soft-tissue window showing abscess surrounding the eroded right condylar process

examination. Meanwhile, the patient was prescribed capsule amoxycillin (with *Lactobacillus*) 500 mg three times a day (T.I.D), tablet metronidazole 400 mg T.I.D and tablet diclofenac sodium 50 mg as needed for 5 days. The swelling worsened and there was no relief in the signs and symptoms after 5 days. The pus smear showed a few epithelioid cells, histiocytes and chronic inflammatory cells (Figure 5), which were suggestive of chronic granulomatous infection.

Based on the cytopathology report along with raised ESR and non-responsiveness towards conventional antibiotic treatment, chronic granulomatous diseases like tuberculosis (TB) or deep fungal infections were suspected, but the culture result for fungal infection was negative. Also, there was no response to systemic antifungal therapy (tablet fluconazole 200 mg day^{-1}). Tuberculous osteomyelitis of the condyle was suspected.

Although there were no specific identifying features to demonstrate TB, the patient belonged to an area where the incidence of TB is higher, hence it was suggested that TB should not be excluded as a diagnosis and could be the cause of the patient's symptoms. However, tuberculous infection was suspected owing to the presence of epithelioid cells and histiocytes in the pus smear. Further, the inefficacy of the conventional antibiotic therapy on the patient cannot be overlooked. Based on these findings, investigations to confirm TB including chest radiograph, Mantoux test, culture and staining of acid fast bacilli (AFB) were advised. All the tests were negative. The value of Mantoux test was 9 mm h^{-1} .

Meanwhile, 10 days after his first visit, the patient complained of pain in the back. The patient was referred for detailed general physical examination. Tenderness in the L5-S1 area and in right sacroiliac joint area was elicited. MRI of the lumbosacral spine and sacroiliac joint was done in axial, coronal and sagittal planes with Signa Profile (GE Medical Systems) 0.2 Tesla MRI

Figure 5 Pus smear showing epithelioid cells. Histiocytes, lymphocytes and red blood cells are also seen

machine. T_2 weighted, T_1 weighted sagittal sequence and coronal short tau inversion recovery sequence was taken.

MRI findings

There was evidence of bilateral paravertebral and pre-vertebral abscess at L5–S1 level (Figure 6) and inflammation and erosion in the right sacroiliac joint, which is highly suggestive of caries of the right sacroiliac joint.

Based on these findings, the final diagnosis of tuberculous osteomyelitis of the right mandibular condyle with caries in the sacroiliac joint was made. The patient was referred for anti-tubercular treatment (ATT) without any delay.

Management

ATT was started. Duration of the treatment was 9 months. Prescription involved first-line anti-tubercular drugs. The patient showed an excellent response to treatment so no consideration was given to surgical intervention. Complete regression of the swelling was achieved within 1 month of ATT (Figure 7). Digital panoramic radiograph (Figure 8) and CT scan (Figure 9) were repeated after 4 months of treatment. The radiographs had no evidence of any abscess or lymphadenopathy. Since the foci of infection had not been traced, the other physicians he had been consulting in the past were approached. On rechecking his medical records, it was found that he was diagnosed as a case of pulmonary TB by a local government hospital 7 years earlier and had been under ATT.

Discussion

TB is one of the oldest diseases known to affect humans. The causative organism, *Mycobacterium tuberculosis*, infects 20–43% of the world's population. It is very important to diagnose this disease at an early stage. If left untreated, it may prove fatal within 5 years in more than half of the cases.¹ There was a massive decrease in the incidence of this disease in 1980s due to advances in chemotherapy, inoculation with BCG (Bacillus Calmette-Guérin) vaccine and improvement in public health and nutritional status. This decline was

Figure 6 Axial T_2 weighted image showing bilateral paravertebral and pre-vertebral collections at L5–S1

Figure 7 Swelling completely regressed after 1 month of treatment

reversed in 1985 owing to population explosion, immigration from developing nations, increase in HIV (human immunodeficiency virus) positive cases and development of multiple drug resistance worldwide.^{1,2}

According to the World Health Organization (WHO) in the late 1990s, between 3.5–4 million new cases of TB were diagnosed annually.¹ These cases represented only a fraction of the total affected people owing to a low level of case detection and incomplete notification in many of the national programmes. The new reported cases rose to 8 million in 1997. In the same year, 2 million patients died of TB, 98% of whom were from developing countries.¹ Without greater control efforts, the annual global incidence of TB may increase by 40% between now and 2020.¹

Although the primary lesion occurs as a pulmonary infection, the extrapulmonary infections have also shown an increase over the past few years. Surprisingly, it is the musculoskeletal system which is more commonly affected than the respiratory system. Most of the cases in the literature regarding TB of jaws are primary. Rarely, secondary oral manifestations associated with pulmonary infection are seen, which can appear as lesions on the gingiva, palate, lips, tongue, buccal mucosa, frenulum and in the jaw bones.⁴

This case deserves special attention as tuberculous osteomyelitis is very rare and constitutes less than 2% of the skeletal TB. Jaw involvement in TB is even rarer and usually affects older individuals.³ Most likely, this case of tuberculous involvement of the condyle with complete lytic pattern along with caries of spine has not been reported so far in dental literature. Besides, it can also be easily misdiagnosed owing to its atypical signs and symptoms and atypical radiographic appearance.

The diagnosis of such cases is based on clinical findings and radiographs, which have to be confirmed by sputum or tissue smears showing the bacilli.^{2,3} The

Figure 8 Panoramic radiograph after 4 months of treatment showing bone formation as compared with previous pre-treatment radiograph

site-specific tissue or fluid aspirate must be submitted for smear culture and histological analysis as has been done in this case. Histologically, it presents as caseating or non-caseating granuloma with giant cells, which was not so in our case. Radiographically, it usually presents as blurring of bone details producing a diffuse radiolucency and erosion of bone, as in this reported case, or it may be a mixed radiolucent-radiopaque lesion.³ About half of such patients do not have pulmonary disease at the time

they report, as the primary infection heals leaving some surviving tubercle bacilli.⁴ With a lowering of resistance, these are reactivated producing local as well as haematogenous spread.^{4,5} TB should be considered in the differential diagnosis of chronic joint disease⁴ even if routine investigations for TB are negative.⁵

A study by Lanka *et al*⁶ on the role of fine needle aspiration cytology of lymph nodes in the diagnosis of cutaneous TB states that the fine needle aspiration cytology smears in their patients with cutaneous TB with lymphadenitis were negative for AFB by the Ziehl-Neelsen stain, while varying degrees of positivity are reported by others. It was concluded that an absence of AFB in smears showing an otherwise characteristic cytological picture should not weigh against the diagnosis of TB.^{7,8} They state that all their patients in the said study responded to standard anti-TB treatment.

Furthermore, according to Pandit *et al*,⁹ when considering the overall prevalence of TB in the Indian population, the presence of epithelioid cell granuloma is indicative of TB unless proven otherwise, which is similar to our findings. It is also reported that the percentage of cases showing AFB positivity declines when more epithelioid cell granulomas are observed.¹⁰

In our country, a standard recommendation of 10 mm is given in the Revised National Tuberculosis Program (RNTPC) guidelines and is both appropriate and relevant for our country. The situation changes in population surveys where a cut off of >15 mm is taken to make the test more specific. Such surveys use Mantoux test as the only criteria to find out the prevalence of tubercular disease without the help of risk factors for any other investigation. But many authors, such as Jorgen¹¹ and Diba *et al*¹², have stated that the cut-off varies from

Figure 9 Coronal CT with contrast in soft-tissue window after 4 months of treatment without any abscess surrounding the resorbed condyle and without lymphadenopathy

region to region depending on the prevalence of atypical mycobacteria and BCG vaccination.

Some authors have also stated that if there is a higher probability of TB infection, such as in the case of recent contact with an infectious case or residence in a high prevalence country as in our case, then an induration of 8 mm or greater is more likely to be due to TB.

Based upon these findings, we conclude that even if the titre of the Mantoux test is borderline, as in this case, the diagnosis and treatment for latent tubercular infection is considered if the person (a) was in contact with another person with infectious TB, (b) was born or has lived in a high TB prevalence country or (c) is continually exposed to populations where TB prevalence is high.

There is no specific standard for diagnosing TB in young patients and under some circumstances diagnosis can be based on circumstantial evidence, which in the present case included a smear-positive TB patient, clinical and radiographic features consistent with TB and a very good response to treatment. Further caries of spine in MRI also added weight towards tuberculous infection.

WHO guidelines for appropriate TB chemotherapy include the necessity for a long-term treatment interval with a multidrug regimen, which can extend up to 9 months.³ Some authors suggest that medical treatment alone is effective. Others believe that aggressive debridement with primary closure is required as an adjunct to chemotherapy to prevent recurrence or the formation of a draining sinus.^{13,14} According to some authors, surgery is not indicated in smaller lesions. Close follow-up is advised in such patients to determine the need for surgical treatment.¹⁴

The role of a history of TB is very important in such cases; however, in this presented case the patient and his

parents denied positive history of the disease. The patient did not consider the disease as significant as he was quite young when he suffered from TB. The parents denied it as they considered it as a social stigma for their son to be seen as a TB patient. So these patients usually try to hide any history regarding TB as in this reported case. Hence, proper counselling of the patient as well as parents is stressed to reveal their medical history, which sometimes proves very beneficial to achieve a correct diagnosis. With varying presentations, the particular disease has a tendency to get overlooked. This report highlights a rare case of TB of the condyle. It is an attempt to familiarize the general dental practitioner on the various presentations of TB and hopefully lead to a reduction in the unnecessary investigations leading to erroneous treatment.

It can be concluded that the morbidity and emergence of TB can be effectively reduced with early diagnosis and initiation of effective regimen. With the increased prevalence of TB, especially in developing countries, the chances of patients reporting first to a dental professional are very high. Leighty *et al*¹⁵ reported that once the bacteria gain access to the synovial space, they can cause irreversible changes within a few days. Thus, the dental professional plays a crucial role in identifying such high risk patients and can initiate prompt isolation and management.

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