Tuberculosis of the temporomandibular joint: A rare case report and review of the literature

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ABSTRACT

Head and neck tuberculosis represents a rare form of extrapulmonary tuberculosis and usually concerns the cervical lymph nodes. Involvement of the temporomandibular joint (TMJ) is extremely rare and only reported at a level of case report forms. A 20-year-old female patient of African origin presented with pain in the mandible and swelling at the left masticator since one month ago. Head and brain CT scan revealed multiple hyperdense lesions with peripheral enhancement and surrounding swelling. These findings were consistent with multiple abscesses. Moreover, severe impairment and the presence of fluid was observed at the left temporomandibular joint, the mandible and the left masseter muscle. Chest CT scan revealed centrilobular nodules in both upper lung lobes. Treatment with common antibiotics was initiated but the patient did not improve. During hospitalization, the patient experienced an episode of generalized tonic-clonic convulsion. Therefore, a surgical drainage of the mandibular abscess was performed. The AFB smear and the molecular test (XPERT MTB/RIF) were positive for *Mycobacterium tuberculosis* sensitive to rifampicin. The patient was treated with isoniazid, high dose rifampicin (900 mg), pyrazinamide, ethambutol and moxifloxacin. The patient received treatment for 12 months and improved significantly clinically and radiologically. Diagnosis of tuberculosis in the temporomandibular joint is challenging but should be included in the differential diagnosis when a compatible history of exposure and other organ involvement exist.

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INTRODUCTION

In 2021, the estimated number of people diagnosed with tuberculosis (TB) increased by 4.5%, mainly due to the disruption of TB services caused by the COVD-19 pandemic leading to delayed diagnosis and treatment initiation. Although specific data regarding extrapulmonary (EPTB) incidence are not available, it is estimated that EPTB accounts for approximately 15–25% of TB infections[1]. Head and neck TB (HNTB) represents 10% of EPTB cases and mainly affects the cervical lymph nodes, larynx, middle ear, oral cavity and pharynx[2,3]. More specifically, TB affecting the orofacial region is very rare affecting only 0.1–5% of extrapulmonary TB cases[4].

Involvement of the temporomandibular joint (TMJ) is extremely rare, with only a few cases reported in the literature. As a matter of fact, TMJ TB accounts for <2% of skeletal TB sites[5].

Unfortunately, symptoms usually appear at an advanced stage and include mainly preauricular swelling, trismus (difficulty in mouth opening) and joint stiffness[6]. The main abnormality is formation of an abscess along with varying degrees of bone destruction, which can result even in the formation of a fistula[4].

Simple imaging studies such as panoramic dental X-rays, show the osteolytic lesion or lysis and condensation usually with fuzzy limits. CT scans provide a more detailed illustration of the abnormalities, including periodontitis with bone loss, bone remodeling with osteolysis and osteo condensation, and pre-mandibular soft tissue abscess[7]. MRI may be more effective in detecting TB abnormalities in peripheral joints. Even though the presence of pus either on CT or MRI is not always apparent, the presence of fluid favors a diagnosis of TMJ TB[8].

The infection begins in the subchondral region and progresses to affect the cartilage, synovium and joint space. The cancellous portion of the mandibular condyle is particularly susceptible to TB, since this part of the bone is affected in the beginning[4].

TMJ TB accounts for an osteoarticular TB form as it usually involves the cancellous portion of the mandibular condyle. *M. tuberculosis* inoculates there by hematogenous dissemination[4]. Nevertheless, TMJ TB can be a complicated manifestation of TB of the oral cavity. Oral TB, although

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uncommon due to natural defensive mechanisms, can involve the buccal mucosa, tongue, gingiva, and mandible[9]. Interestingly, in an older review concerning TB of the oral cavity, it was reported that the second most commonly affected area was the mandible (21.4%)[10]. Mandibular osteomyelitis presents with signs of atypical osteitis, periodontal disease with vertical bone loss, or destructive osteolysis. Discharge of pus or blood was described either by sinuses, tooth sockets, even by the spicules of bones through the gums[10]. It appears that abnormalities follow gradual involvement of adjacent tissues but it is not known which is the primary site of infection; the mandible or is it the oral cavity? The latter can be explained as it is known that breaks in the continuity of the mucosa, such as trauma, allow invasion of *M. tuberculosis* into deeper tissues, including the facial bones[9]. Local factors that may facilitate invasion of the oral mucosa include poor oral hygiene, leukoplakia, local trauma, and irritation by clove chewing[11]. Interestingly, TMJ TB has even been reported to arise from a fistulous communication from the middle ear[12].

Diagnosing TMJ TB requires confirming the presence of *M. tuberculosis* through microbiological tests such as AFB, molecular or culture, either by bone biopsy or more commonly via a fine needle aspiration of the pus. While TMJ TB can rarely present as a primary form of TB[4], it is more commonly associated with pulmonary disease. When a patient presents with both pulmonary TB and TMJ abnormalities, it is highly likely that TMJ TB is also present. In this case, a good response to treatment, both clinically and based on imaging findings is expected.

Differential diagnosis for TMJ TB includes other conditions like abscess, arthritis, osteomyelitis, neoplasmatic disease, or any systemic diseases that affect the joints. Pathology of the surrounding structures, e.g. teeth and the parotid gland, have to be ruled out.

Treatment of TMJ TB is similar to that of pulmonary TB, although it requires a significant amount of time, typically 6–9 months or even longer. The presence of other affected organs from tuberculosis also determines the final regimen. In our case, which is the first reported to have concomitant pulmonary and CNS tuberculosis, a high dose of rifampicin was used (900 mg), given new data. Moreover, since *M. tuberculosis* was sensitive to all first-class medicines a quinolone was added to increase treatment efficacy regarding CNS involvement. The extent of TMJ destruction at the time of diagnosis and treatment initiation determines the level of functionality afterwards. Early diagnosis is crucial for achieving the best possible quality of life. In fact, around 90–95% of TMJ TB patients regain normal masticatory function, when treatment is initiated at an early stage[13]. Surgical interventions such as decortications and excision are only considered when oral drug treatment seems insufficient[4]. The treatment of orofacial TB varies, depending on its presentation. According to the literature, cases with minimal destructive lesions typically respond to medical intervention with anti-tubercular therapy, leading to resolution, without the need for surgical treatment. On the other hand, moderately destructive lesions may necessitate decortication of bone due to medullary bone destruction and/or cortical bone perforation[14].

CASE PRESENTATION

A 20-year-old female patient of African origin presented to the hospital with pain in the mandible, swelling at the left masticator and inability of complete mouth opening since one month ago. The patient was admitted to the internal medicine department for further investigation and treatment.

The patient had no significant medical history, except for a tooth extraction 3 years ago, and was not taking any medications. She was afebrile, and all her vital signs were within the normal range. Swelling and pain at palpation were observed upon palpation of the left temporomandibular joint and the masticator muscle area. Additionally, the patient had proptosis (exophthalmos) in her right eye without experiencing diplopia. She mentioned a non-productive cough over the past month. Neurologic examination revealed no deficits.

The patient underwent a chest and brain CT scan which revealed multiple hyperdense lesions in the right frontal and temporal lobe with peripheral enhancement and surrounding swelling. The largest lesion was 1.6 cm in the frontal lobe (Figure 1). Leptomeningeal enhancement was also observed. These findings were consistent with multiple abscesses and meningitis. Moreover, severe impairment and the presence of an abscess were observed in the right parietal bone. The

Figure 1. CT brain: hyperdense lesion in the right frontal lobe (1.6 cm) with peripheral enhancement and surrounding swelling

Table 1. Overview of cases of TB infections of the TMJ

Authors	Age	Gender	Symptoms/ clinical findings	Imaging findings	Type of operation	Other sites of TB co-infection	Type and duration of treatment
Geetha et al.[4]	49	Female	Swelling in front of the left tragus	Destruction of the left mandibular condyle with proximal sclerosis and erosion of mandibular foss	Condylectomy	None	6 months standard regimen
Helbling et al.[6]	22	Female	Swelling and pain in the left preauricular area	Destruction of the left condyle and condylar fossa	FNA	None	9 months standard regimen
Park et al.[8]	53	Male	Painful swelling in the right preauricular area + difficulty in mouth opening since 3 months	Destructive changes of the right condyle glenoid fossa and ring enhancement of the preauricular area of the TMJ	Incision and drainage	Lumbar spondylodiscitis	6 months standard regimen
Mohad et al.[15]	12	Male	Swelling in the right preauricular area and difficulty in mouth opening since 2 months	Pronounced rarefaction and destruction of bone in mandibular condyle with discontinuity of the cortical boundary	FNA + incision	None	Unknown duration, standard regimen
Sheikh et al.[16]	20	Male	Swelling in the right preauricular area	Erosion with comminuted destruction of the right mandibular condyle + abscess	Us guided FNA	Bilateral paravertebral and prevertebral abscess at L5–S1 level + right sacroiliac joint	9 months standard regimen
Kumar et al.[17]	35	Male	Swelling in the left preauricular area + difficulty in mouth opening	Erosion with the trabecular destruction of the left mandibular condyle + abscess+ cervical lymphadenopathy	FNA	None	INH/RIF for 9 months + condylectomy
Koul et al.[18]	16	Female	Left-sided preauricular facial swelling + trismus	Rarefaction + destruction of bone with a large mass in retromandibular and inferior temporal fossa	Trucut biopsy	None	9 months standard regimen
Karjodkar et al.[19]	A 18	Female	Swelling on left preauricular region + trismus since 8 months	Osteomyelitic changes in relation to left condyle and ramus	FNA	None	NA
	B 45	Male	Swelling on right side at the angle of the jaw since 2 months + teeth extraction due to pain	Irregular destruction at the angle region + osteomyelitis of the mandible	FNA	None	NA

Figure 2. CT brain: destruction of the tuber of the left mandible, the temporal bone and edema of the left masseter muscle

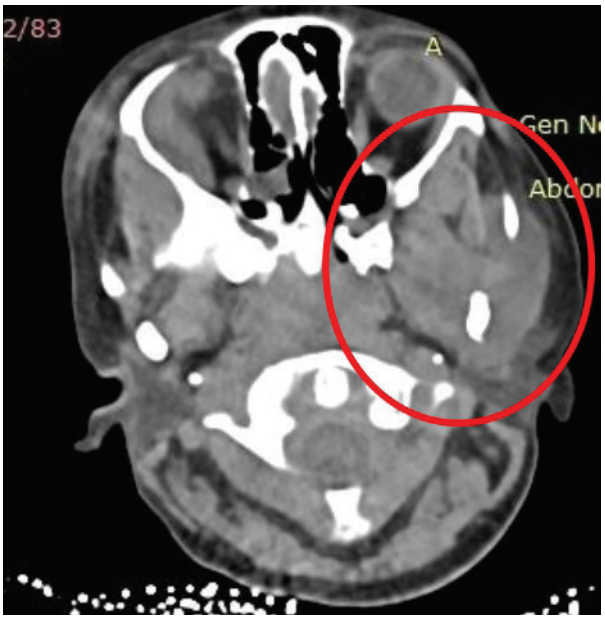
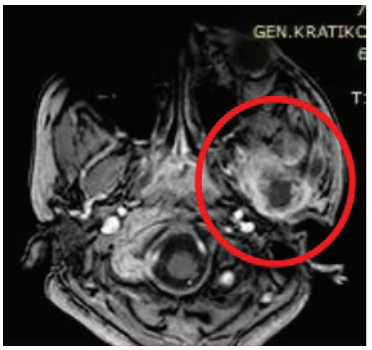


Figure 3. Brain MRI: abscess and edema surrounding at the left temporomandibular joint



tuber of the left mandible, the mandible itself, as well as the left masseter muscle appeared to be affected (Figures 2 and 3). Fluid consistent with an abscess was also present at the area. Since MRI is more sensitive than CT scanning in determining the extent of meningeal and parenchymal involvement, an MRI was performed and confirmed all the aforementioned lesions. Chest CT scan revealed centrilobular nodules and traction bronchiectasis in both upper lung lobes.

Laboratory tests showed elevated inflammatory markers (CRP, ESR). The patient tested negative for HIV, HCV and HBV, while the Quantiferon test was positive.

Initially, the patient received common antibiotics with ceftriaxone and vancomycin but showed no improvement. Therefore, surgical drainage of the mandibular abscess was performed. The drained fluid was examined for common bacteria, fungus, toxoplasma, nocardia and mycobacteria. The AFB smear and the molecular test (XPERT MTB/RIF) were positive for M. tuberculosis. The detected strain was sensitive to rifampicin.

The patient was treated with isoniazid, high dose rifampicin (900 mg), pyrazinamide, ethambutol, and moxifloxacin. Current data support the use of higher doses of rifampicin (i.e. 35 mg/kg) in order to achieve increased CNS penetration. The use of levofloxacin instead of moxifloxacin is preferable due to reduced levels of the latter when co-administered with rifampicin. However, the patient presented an allergic reaction to levofloxacin, and moxifloxacin was administered instead. The patient responded well to the treatment both clinically and radiologically, in terms of CT findings.

During hospitalization, the patient experienced an episode of generalized tonic-clonic convulsions, resulting in a traumatic fracture of the cervical vertebrae A2. A brain CT scan was repeated and revealed further damage of the odontoid process of the A2 vertebrae. The patient was started on daily anticonvulsant treatment (levetiracetam) and no further seizures occurred.

The patient was discharged from the hospital after 34 days and continued follow-up at the Antituberculous Department of the Chest Disease Hospital. She received treatment for another 12 months and improved significantly. Follow-up CT scans showed improvement or remission of most lesions in both the brain and chest.

DISCUSSION

Diagnosis of TMJ TB can be challenging, especially when it presents as the primary symptom. Delayed diagnosis may be observed due to a lack of strong suspicion. Patients often lack pathognomonic signs, and exhibit fewer systemic symptoms, and even the typical tests performed when TB is suspected such as Tuberculin Skin Test (TST) yield positive results in only about half of the patients (53%)[14]. Unlike pulmonary TB, only 20% of head and neck TB patients experience typical symptoms such as cough, fever, or night sweats[14]. An overview of relevant studies of cases of TB infections of the TMJ is given in Table 1.

CONCLUSION

This case presentation emphasizes the importance of considering TB as part of the differential diagnosis, particularly for patients from endemic areas. In such cases investigation for lung involvement reinforces the diagnosis. Regrettably, there is a growing necessity to report and record each case of extrapulmonary TB, in order to guide the diagnostic approach for uncommon sites of infection.

CONFLICTS OF INTEREST

The authors have completed and submitted the ICMJE Form for disclosure of Potential Conflicts of Interest and none was reported.

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ETHICAL APPROVAL AND INFORMED CONSENT

Ethical approval and informed consent were not required for this study.

DATA AVAILABILITY

The data supporting this research are available from the authors on reasonable request.

PROVENANCE AND PEER REVIEW

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