

# Tuberculosis of the Temporomandibular Joint: A Case of Misdiagnosis

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Tuberculosis (TB), an infectious disease caused by *Mycobacterium tuberculosis*, continues to be a major health problem worldwide. Primary TB infection is mostly pulmonary, but it may also occur in the lymph nodes, abdomen, skin, meninges, joints, and central nervous system. TB in the head and neck region usually occurs in the cervical lymph nodes, but is extremely rare in the temporomandibular joint (TMJ). This rarity increases the risk for not considering TB in the differential diagnosis of patients presenting with TMJ pain. This article describes an unusual case of a 53-year-old man with painful swelling in the right preauricular area accompanied by difficulty in mouth opening. After surgical exploration, histopathologic findings revealed TB in the TMJ. The findings of this case highlight the importance of considering TB in the differential diagnosis of TMJ pain, especially for patients from endemic areas, patients who have lived or visited those areas, or patients with a preauricular swelling in the TMJ area. *J Oral Facial Pain Headache* 2014;28: 165–170. doi: 10.11607/ofph.1061

**Key words:** *extrapulmonary tuberculosis, preauricular swelling, temporomandibular joint, temporomandibular disorders*

**T**uberculosis (TB), an infectious disease caused by *Mycobacterium tuberculosis*, has recently become the second leading cause of death from infectious diseases worldwide.<sup>1</sup> It affects mainly the lungs (pulmonary TB), but can also affect other organs (extrapulmonary TB), such as pleura, lymph nodes, abdomen, genitourinary tract, skin, joints, bone, and meninges.<sup>1,2</sup> The diagnosis of an extrapulmonary TB can represent a clinical challenge in the absence of the classic symptoms associated with pulmonary TB.

According to the World Health Organization, in 2010 4.6 million of people who were diagnosed with TB for the first time (new cases) had pulmonary TB and 0.8 million had extrapulmonary TB.<sup>1</sup> The prevalence of extrapulmonary TB has increased in the last 2 years, especially as a consequence of the HIV epidemic,<sup>3</sup> and this form is found in more than 50% of HIV-positive patients with TB.<sup>4</sup> Fifty-nine percent of the new TB cases worldwide occurred in Asia, especially in China and India. The incidence of TB in South Korea also continues to increase; in 2010 it was 97 cases per 100,000 people, while in China it was 78 cases per 100,000 people, leaving the former country with one of the highest TB burdens worldwide.<sup>1</sup>

About 10% of all extrapulmonary TB present in the head and neck, with the cervical lymph nodes being the most commonly affected site.<sup>5</sup> TB infections of the facial bones have also been described.<sup>6–8</sup> The temporomandibular joint (TMJ) is rarely involved and only a few cases have been reported in the literature (Table 1), most of them classified as extrapulmonary TB.<sup>2,9–12</sup> It has been estimated that in India, which has about 26% of all TB cases worldwide,<sup>1</sup> only 1 out of 165 cases of TB involving the head and neck region exhibited a TMJ involvement.<sup>5</sup> This rarity increases the risk for TB not being considered in the differential diagnosis of patients presenting with TMJ pain. This article presents a case of a patient with TB in the TMJ and draws attention to the diagnostic pitfalls in such a case of a rare pathology of the TMJ.

**Table 1** Reported Cases of TB Involving the TMJ

	Age (years)	Sex	Side	Symptoms	Diagnosis
Helbling et al <sup>2</sup>	22	Female	Left	Swelling, pain	Biopsy, FNAC
Wu et al <sup>9</sup>	59	Female	Left	Swelling	Biopsy
Coscaron Blanco et al <sup>10</sup>	30	Female	Left	Swelling, pain, fever	FNAC
Ruggiero et al <sup>11</sup>	22	Female	Left	Swelling, difficulty opening mouth	Culture for <i>Mycobacterium</i>
Present case	53	Male	Right	Swelling, pain, difficulty in opening mouth	Biopsy

FNAC, five-needle aspiration cytology.

## Case Presentation

### History

A 53-year-old man was referred to the Department of Oral and Maxillofacial Surgery from the Department of Oral Medicine, College of Dentistry, Yonsei University, for the evaluation of right preauricular painful swelling and difficulty in mouth opening. The pain had started spontaneously 3 months prior to the referral and was accompanied by a progressively increasing difficulty in mouth opening. At that time there was no swelling of the TMJ area.

The patient had contacted two specialists in temporomandibular disorders (TMD) at two local dental clinics, who diagnosed osteoarthritis of the right TMJ and treated him with an occlusal splint and nonsteroidal anti-inflammatory drugs, but without success. Meanwhile, the patient began experiencing a progressively increasing preauricular swelling on the right side that led to facial asymmetry.

The patient was otherwise in good health. He had no known drug allergies, was not regularly taking medications, and could not recall having had any symptoms of TB, such as chronic cough with blood-tinged sputum, fever, night sweats, or weight loss. He also did not recall a history of trauma or recent dental treatment, except for a thoracic-spine operation due to a herniated intervertebral disc 8 years previously.

### Clinical Examination and Imaging

At the initial visit at the department, the patient complained of difficulty in opening the mouth and of sharp pain radiating from the right side of the face to the neck and the back of the head that exacerbated during mouth opening and mastication.

The extraoral examination revealed a 1.5 × 1.5-cm preauricular firm and tender swelling on the right side accompanied by redness and a temperature increase of the skin. Palpation of the cervical lymph nodes was negative. The active maximum mouth opening was reduced to 22 mm. Lateral excursions were 7 mm to the right and 4 mm to the left, without pain. Protrusion reached 4 mm, without pain, but with some deviation to the right. Palpation of the masticatory muscle was negative. The intraoral examination did not reveal any abnormalities of the floor of the mouth, the tongue, the buccal mucosa or palate. Body temperature and

vital signs were within normal limits. The initial laboratory tests showed a white cell count of 8,510 cells/mm<sup>3</sup> with a normal differential count and an increased erythrocyte sedimentation rate of 85 mm for the first hour (normal value, 14 mm/h).

The initial clinical differential diagnosis included benign and malignant neoplasms, such as osteochondroma, synovial chondromatosis, osteosarcoma, or chondrosarcoma; TMJ arthritis; a TMJ infection such as a septic arthritis with a local or systemic cause; or parotitis.

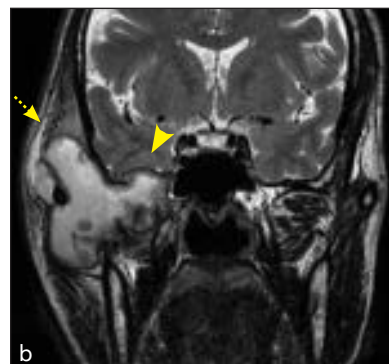
The panoramic and cephalometric posterior-anterior views performed in the department did not show pathologic bony alterations caused by an odontogenic infection. Because of the preauricular swelling, magnetic resonance imaging (MRI) and computed tomography (CT) contrast-enhanced imaging were requested. The MRI revealed a scalloped ring area of high-signal intensity surrounding a low-signal area around the right TMJ, suggesting the presence of fluid or exudate. The scalloped ring extended into the right infratemporal fossa, temporal fossa, masticatory spaces, and the right middle cranial fossa with dural thickening (Fig 1). The contrast-enhanced CT scans disclosed destructive changes of the right condyle, glenoid fossa, middle cranial fossa, and around the foramen ovale, and ring enhancement in the preauricular area of the right TMJ (Fig 2).

Based on these findings it was decided to perform an intraoral and extraoral incision and drainage. The intraoral incision was performed along the ascending ramus, while the extraoral incision was made in the right preauricular and temporal areas. Silastic drains were inserted in both incision sites and the area was irrigated daily with normal saline solution. Yellowish pus flowed through both drains for 2 weeks. The drains were removed 1 week later. The culture of the material obtained intraorally revealed the presence of methicillin-sensitive *Staphylococcus aureus*, and that of the first culture of the extraorally collected pus *Pseudomonas aeruginosa* and *Klebsiella oxytoca*. The second culture of the extraorally obtained material contained *Stenotrophomonas* species. This led to the diagnosis of TMJ septic arthritis, the source of the infection remaining unknown. None of the bacterial cultures contained *Mycobacterium* species. The patient was therefore treated medically with antibiotics (ubacillin 3 g/day).

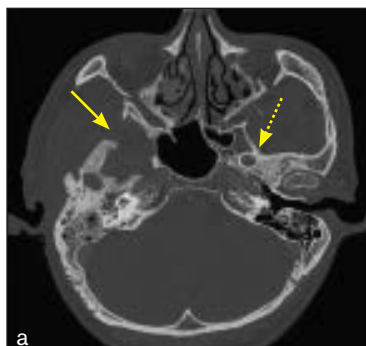
**Fig 1a** Axial gadolinium-based contrast-enhanced T1-weighted MRI image showing a high-intensity scalloped ring (*arrow*) surrounding a low-intensity area in and around the right.



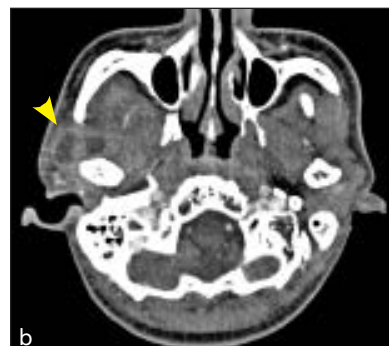
**Fig 1b** Coronal contrast-enhanced T2-weighted MRI image showing a high-intensity area extending to the right middle cranial fossa (*arrowhead*) and temporal space (*dotted arrow*).



**Fig 2a** Axial contrast-enhanced CT image (bone window) showing destruction of the right TMJ, middle cranial fossa, and the area around the foramen ovale (*arrow*). Note the normal left foramen ovale (*dotted arrow*).

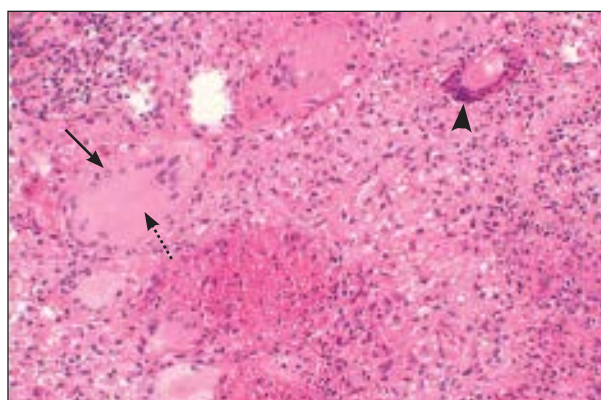


**Fig 2b** Axial contrast-enhanced CT image (soft tissue) showing ring enhancement in the preauricular area of the right TMJ (*arrowhead*).



Meanwhile, the patient started reporting severe low-back pain with a similar onset to that in the TMJ area. He was transferred to the Department of Neurosurgery for evaluation and treatment of the low-back pain. Lumbar spine MRI revealed a diffuse bulging disc at the fifth lumbar/first sacral vertebrae with signal enhancement in the peripheral disc area and endplate. This finding led to an initial diagnosis of spondylodiscitis.

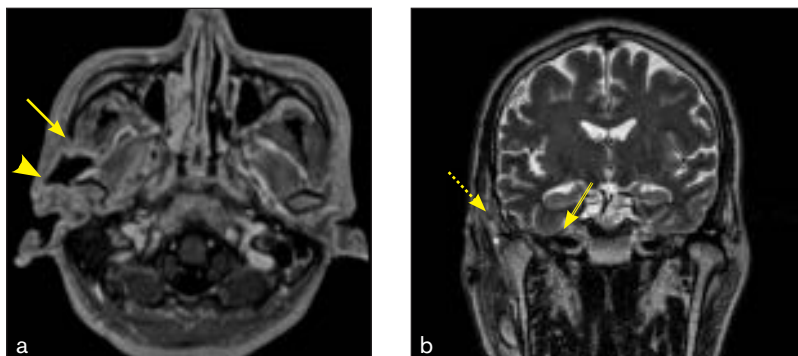
As the condition did not improve under antibiotics, a surgical exploration of the TMJ area was undertaken under general anesthesia. This revealed necrotic tissue extending to the infratemporal space, both superficial and deep temporal spaces, and the area between the medial side of the ramus of the mandible and the lateral surface of the medial pterygoid muscle. The necrotic tissue was debrided while preserving the right condylar head as much as possible to prevent facial asymmetry. Drain tubes were inserted intraorally and extraorally. The histologic evaluation of the necrotic tissue revealed a chronic granulomatous inflammation with caseous necrosis surrounded by epithelioid cells, Langerhans giant cells, and lymphocytes consistent with TB (Fig 3). In order to rule out TB of the spine or another extrapulmonary pathology, positron-emission tomography/CT (PET-CT), a chest CT, acid-fast bacilli (AFB) sputum smear, sputum mycobacterial culture, and HIV tests were requested. The sputum mycobacterial culture was positive for *Mycobacterium tuberculosis*, while the three AFB sputum tests and the HIV test were negative, and the



**Fig 3** Histologic evaluation revealed chronic granulomatous inflammation with caseous necrosis (*dotted arrow*) surrounded by epithelioid cells (*arrow*), multinucleated giant cells (*arrowhead*), and lymphocytes.

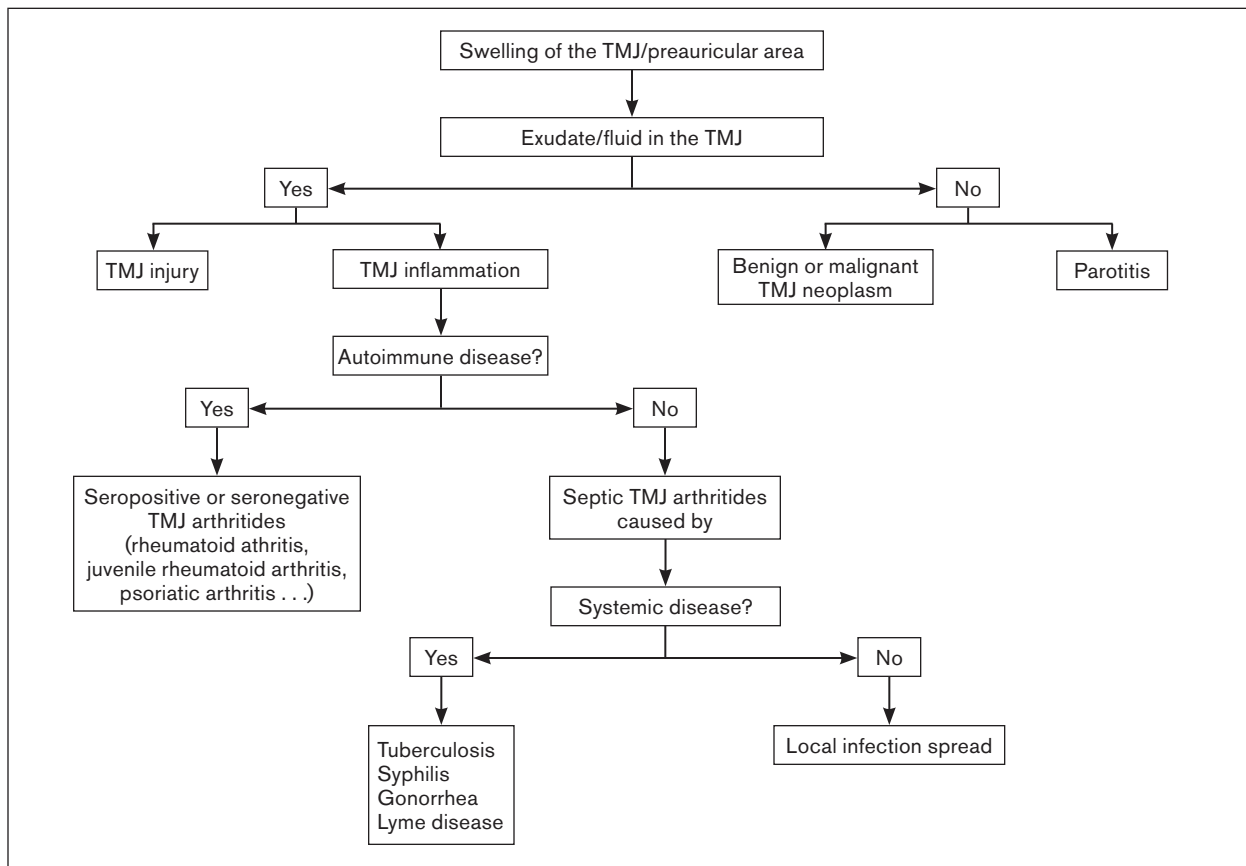
chest X-rays normal. This finding led to the final diagnosis of extrapulmonary TB of the right TMJ and TB spondylodiscitis.

The patient underwent 6 months of chemotherapy involving a combination of rifampicin (600 mg/day); isoniazid (300 mg/day); pyrazinamide (500 mg/day); and ethambutol (400 mg/day), after which all of the symptoms improved. Postoperative MRI revealed a decrease in the extent of the bony lesions and high-intensity area (Fig 4).



**Fig 4a** Axial gadolinium-based contrast-enhanced T1-weighted postoperative MRI showing a reduction in the extent of the lesion around the right TMJ (arrow). The drain insertion site is indicated by an arrowhead.

**Fig 4b** Coronal T2-weighted postoperative MRI showing reduced pathologic enhancement in the right temporal space (dotted arrow) and middle cranial fossa (double-stranded arrow).



**Fig 5** Diagnostic flow chart for a swollen TMJ.

## Discussion

Primary TB infections are the result of direct person-to-person spread through air-borne droplets from a patient with active disease. Primary TB occurs in previously unexposed people and almost always involves the lungs. The infection is usually asymptomatic, but occasionally it is accompanied by fever and pleural effusion. Extrapulmonary TB is usually caused by hematogenous dissemination of the Mycobacterium,<sup>2</sup> and joint involvement appears by hematogenous spread leading to a bacterial colo-

nization of the vascular cancellous bone, also known as osteoarticular TB.<sup>13</sup> Osteoarticular TB usually occurs in the spine (approximately 50%) as well as in the metaphysis and epiphysis of long bones.<sup>14</sup> TB of the TMJ can be considered as an osteoarticular form of TB due to its tendency to show tubercular involvement in the cancellous portion of the mandibular condyle.<sup>14</sup>

TB of the TMJ is characterized by painful preauricular swelling that is usually unresponsive to antibiotic therapy and is associated with difficulty in mouth opening.<sup>2,9-12</sup> The differential diagnosis in the



case of swelling in the preauricular area includes TMJ pathologies, such as inflammation or infection and benign and malignant neoplasms, but also pathologies of the surrounding structures, eg, of the teeth, ear, and parotid gland (Fig 5). Septic TMJ arthritis can be caused by hematogenous spread or by an infection of the surrounding area, such as an odontogenic infection spreading to the TMJ. This condition should be easy to diagnose by imaging of the dental arches and intraoral examination, although in cases of severe trismus this may be difficult. In the absence of a local infection, hematogenous spread from a distant source or systemic diseases (such as tuberculosis, syphilis, and gonorrhea) must be considered.<sup>15</sup> In the present case, there were no signs of a possible odontogenic infection of the right TMJ so a systemic origin had to be considered.

The final diagnosis of osteoarticular TB was delayed because the source of infection could not be found, which makes this case report significant. Indeed, the diagnosis of extrapulmonary TB represents a clinical challenge when the classic symptoms associated with pulmonary TB, such as fever, cough, weight loss, and night sweats, are not present.<sup>13</sup> Differential diagnostic tests for TB should be considered during the initial diagnostic process of a TMJ infection, especially in areas in which TB is prevalent or for people who have lived or visited such areas. These tests include cultures of *Mycobacterium tuberculosis* from bone tissue, AFB sputum smear, fine-needle aspiration cytology (FNAC), and surgical biopsy.<sup>14</sup> FNAC and biopsy usually confirm the diagnosis of a TB infection, if present. In this case, none of the extraoral and intraoral bacterial cultures contained *Mycobacterium*, the reason being that because TB was not considered in the differential diagnosis, the pus cultures were performed with a conventional medium (agarose gel) and not the solid and broth mediums necessary for the *Mycobacterium* culture. The presence of *Mycobacterium* was observed only in the sputum culture. This explains why the final diagnosis, confirmed with histopathology, was delayed.

The therapy of extrapulmonary TB is identical to that of the pulmonary form. The treatment of osteoarticular TB requires prolonged antibiotic administration.<sup>2</sup> With early treatment, approximately 90% to 95% of patients can be cured without any development of masticatory function impairment.<sup>16</sup> Surgical excision and decortications are indicated only for cases refractory to the antibiotic drug therapy.<sup>2</sup> The functional prognosis of the joint depends on the degree of destruction at the time of the correct diagnosis and the start of treatment. This underlines the need for early diagnosis, as a delayed or missed diagnosis can result in severe destruction of the TMJ that leads to significant functional problems.<sup>14</sup>

It has been proposed that MRI imaging improves the clinician's ability to make the differential diagnosis.<sup>17</sup> The MRI features of peripheral joints infected by TB are bone resorption and areas of high-intensity signal in the joint and its surrounding region that depict the presence of fluid.<sup>17</sup> It is difficult to diagnose the presence of pus from a CT or MRI. Nonetheless, it has been shown that a ring enhancement and particularly an irregularity (scalloping) of the collection wall in the CT images are indicators of the presence of pus in the affected area.<sup>18</sup> For instance, while a ring enhancement has a high sensitivity of 89% and a specificity of 0%, the presence of scalloping yields a sensitivity of 64%, a specificity of 82%, and a positive predictive value of 94%.<sup>18</sup> The CT scans in the present case revealed scalloping and a ring enhancement around the right TMJ that suggested the presence of pus. The flowing pus confirmed the presence of TMJ septic arthritis.

This case report underlines how easy a correct diagnosis of a seldom-occurring TMJ pathology can be missed. TMJ disorders may have several causes, including disc displacement, degenerative disorders, arthritides, systemic diseases, benign and malignant neoplasms, and congenital and development disorders.<sup>19</sup> The first three are the most frequent causes.<sup>20</sup> The fact that these different pathologies often present with the same signs and symptoms, as well as the fact that systemic diseases with manifestation in the TMJs, neoplasms, and congenital and developmental abnormalities are rare, increase the risk of misdiagnosis. Indeed, in the differential diagnostic process, the clinician has the tendency to favor pathologies that occur more frequently. The risk is particularly high when the masticatory symptoms suggest a TMJ disorder but the clinical examination does not reveal swelling of the preauricular area. The risk to diagnose an arthralgia or osteoarthritis in these cases is particularly high. It has been postulated that the low frequency of TB of the TMJ reported in the literature is the consequence of misdiagnosing this condition as osteoarthritis.<sup>17</sup>

## Conclusions

This case emphasizes the need during the differential diagnostic process to take into account rarely occurring pathologies. It also demonstrates that TB of the TMJ should be considered especially in areas with a high TB prevalence, in patients who have lived or visited those areas, and in those with unusual presentations of preauricular swelling accompanied by difficulty in mouth opening. A careful comparison of the patient's history with signs and symptoms is necessary to rule in or out these rare pathologies so as to arrive at a correct diagnosis as early as possible.

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The authors report no conflicts of interest related to this study.

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