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## **Case Report**

# An ambiguous presentation of orofacial tuberculosis – A case report and review

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## Abstract

Tuberculosis is a prevalent systemic bacterial disease caused by *Mycobacterium tuberculosis*. Tuberculous lymphadenitis remains the most common extra pulmonary manifestation of tuberculosis. Tuberculosis in the head and neck area can be varied and often misleading. We present a case of a 10 year old female patient with a recurrent swelling in her left submandibular region. She was diagnosed with tuberculous lymphadenitis based on histopathology report. Tuberculosis in the head and neck is not such a rare disease, but devoid of characteristic clinical and radiographic features, become a diagnostic challenge. Early diagnosis and prompt treatment can thwart complications. Clinicians should be aware of the condition and take it into account in their differential diagnosis.

Keywords: Oral tuberculosis, Tuberculous abscess, Extrapulmonary tuberculosis, Tuberculous lymphadenitis, Orofacial tuberculosis.

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## 1. Introduction

Tuberculosis (TB) is a chronic granulomatous infectious bacterial disease chiefly caused by Mycobacterium tuberculosis.1 Clinical manifestations of tuberculosis are of two types: Pulmonary and Extra Pulmonary.<sup>2</sup> Tuberculosis chiefly involves the pulmonary system which is the lungs and extra pulmonary infection sites include pleura, central nervous system, lymphatic system, genitourinary system, bones and joints.<sup>3</sup> In recent years, numerous extra pulmonary TB cases were reported most commonly in children which frequently involved the head and neck region. The common manifestation in these cases was usually a mass in the cervical region.<sup>1</sup> Clinical features of head and neck tuberculosis in the early stages may simulate neoplasms or other inflammatory diseases as their systemic symptoms are mostly not very obvious then. Therefore clinical diagnosis of TB is usually considered only after an ineffective antiinflammatory treatment, biopsy or sometimes following a surgical resection.<sup>4</sup> Tuberculous lymphadenitis is the lymph node component of primary complex of the oral cavity where gingiva, tongue, and buccal mucosa can be infected.<sup>1</sup> As such

it is considered as an occupational risk for dentists since they work in close proximity to the nasal and oral cavities of these patients who can easily transmit the infection<sup>1</sup>. Cervical group of lymph nodes were further divided into submental, submandibular, postauricular, jugulo-omohyoid, jugulodigastric, and supraclavicular lymph nodes.<sup>5</sup> But a high possibility of suspicion is needed for diagnosing cases of tuberculous lymphadenitis as it can mimic several other pathological conditions.<sup>3</sup> Hence all dentists should equip themselves and understand about the signs and symptoms of head and neck TB for early diagnosis and management of the same.

## 2. Case Report

A 10 year old female patient reported to the department with a chief complaint of recurrent swelling on the left side of her lower jaw since 6 months. Patient also gave a history of incision and drainage along with antibiotic treatment for the past 5 months in the same area.

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On general examination, patient was moderately built and nourished and her past medical & family history were insignificant.

On extra oral examination a well-defined swelling was observed on the left side of her lower jaw along the angle of mandible (**Figure 1**). On inspection, it appeared ovoid in shape and measured about 3\*1.8 cm in diameter. Superimposing skin on the swelling was slightly erythematous compared to the surrounding area. There was no pus discharge noted during inspection. However an extra oral scar adjacent to the swelling was evident on the left premolar region due to the recurrent drainage of pus earlier.



**Figure 1**: A well-defined swelling on the left side of lower jaw along the angle of mandible with an extra oral scar infront of it

On palpation, the swelling was firm and immobile with the central area showing tenderness and fluctuance without any pus discharge. Submandibular and jugulodigastric lymph nodes of the same side were enlarged, firm, tender and nonfluctuant.

On intra oral examination, soft tissue appeared to be normal. Grossly decayed 74 and 75 were noted. A provisional diagnosis of Suppurative Osteomyelitis was initially given based on the above findings. Antibioma and left submandibular sialadenitis were considered as its differential diagnoses. Incision and drainage were done extra orally for the swelling (**Figure 2**).

Patient was then advised for a panoramic radiograph (OPG). OPG only showed grossly decayed 74 and 75 which were then extracted (**Figure 3**). Patient was then advised to take a CBCT and an MRI for further evaluation.

MRI revealed a well defined thick walled subcutaneous abscess measuring about 2.4\* 2 cm on the left submandibular region. Minimal cortical bone irregularity was detected in the inner surface of the left angle of mandible. This was confirmed with the CT scan which also showed a focal bulge of periosteum (**Figure 4**). Multiple enlarged submandibular and jugulodigastric nodes were also reported.



Figure 2: Incision and drainage done extra orally



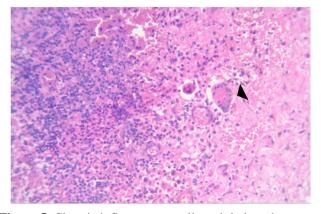
Figure 3: OPG showing grossly decayed 74 and 75



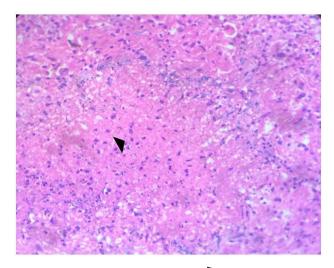
**Figure 4**: CT showing a well-defined thick walled subcutaneous abscess with minimal cortical bone irregularity in the inner surface of the left angle of mandible. Focal bulge of periosteum was also noted

Following the above radiographic examinations, FNAC was performed which yielded a very thick purulent material consisting of dense inflammatory infiltrate mainly neutrophils along with necrosis. Hence FNAC report was given as Suppurative Lymphadenitis.

Finally an incisional biopsy from the swelling was done and six bits of soft tissue specimen were received that were whitish in colour and the biggest bit measured about 2\*0.5 \*0.2 cm. All bits were processed. On microscopic examination, Haematoxylin and Eosin (H &E) stained sections showed wide areas of caseous necrosis with surrounding chronic inflammatory cells chiefly lymphocytes, plasma cells, macrophages and occasionally scattered epitheloid cells at the periphery along with Langhans giant cells. There were also areas showing abscess with clumps of neutrophils. Histopathological features were suggestive of Tuberculous Lymphadenitis with abscess (**Figure 5** and **Figure 6**). Patient was ultimately referred to the physician for further treatment.



**Figure 5**: Chronic inflammatory cells mainly lymphocytes, plasma cells, macrophages and scattered epitheloid cells at the periphery along with Langhans giant cells ( $\checkmark$ ) (H&E, 40x)



**Figure 6**: Areas of caseous necrosis ( $\checkmark$ ) with surrounding chronic inflammatory cells and scattered epitheloid cells at the periphery along with Langhans giant cells. (H&E, 40x)

#### 3. Discussion

The most common form of extrapulmonary TB is tuberculous lymphadenitis also known as scrofula, commonly caused by Mycobacterium tuberculosis. Atypical mycobacterium such as Mycobacterium avium and Mycobacterium kansasii, have also been reported to have caused the above, especially involving children.<sup>6</sup>

Orofacial TB is the rare manifestation of extrapulmonary TB with nonspecific clinical features. Diagnosis becomes even more challenging especially when oral lesions appear before systemic symptoms. Orofacial TB can be manifested in different forms such as tuberculous ulcer, tuberculous gingivitis, tuberculous dental periapical granuloma, tuberculous involvement of extraction sockets of teeth, tuberculous osteomyelitis of jaw, tuberculosis of maxillary sinus, tuberculous of temporomandibular joint, tuberculous sialadenitis, tuberculous lymphadenitis and lupus vulgaris.<sup>7</sup>

In the present case, only a well-defined swelling was observed on the left side of the patient's lower jaw in the region of the angle of mandible along with submandibular and jugulodigastric lymph node enlargement. An OPG that was taken also showed only grossly decayed teeth intra orally corresponding to the same area.

The progression of tuberculous lymphadenitis has been described by Jones and Campbell.<sup>7</sup>

**Table 1:** Stages of progression of tuberculous lymphadenitis jones and campbell classification

Stage 1	Enlarged, firm, mobile, discrete nodes showing
	non specific reactive hyperplasia.
Stage 2	Large rubbery nodes fixed to surrounding
	tissue owing to periosteum
Stage 3	Central softening due to abscess formation
Stage 4	Collar stud abscess formation
Stage 5	Sinus tract formation

In our case, the swelling on the left side of the jaw appeared firm and immobile with the central area showing tenderness and fluctuation without any pus discharge. Hence based on the above classification, patient in the present case belongs to Stage 3.

Orofacial TB is rare and seen in different locations such as the mandible, lymph nodes, tonsils, salivary glands, maxilla/maxillary antrum, hard palate and soft tissues such as soft palate, uvula, gingiva, tongue and buccal mucosa. Oral TB lesions are supposed to be caused by transmission of infected sputum through a break in the mucosal surface during coughing episodes. One of the most common clinical manifestations of oral TB is a non-healing ulcer. Oral TB ulcers usually have an indurated, irregular and undermined margin with a necrotic base.<sup>8</sup>

In the present case, only grossly decayed teeth in the area of swelling were noted and there were no intra oral ulcers resembling TB were seen.

Cervical lymph nodes get involved in tuberculosis due to the entry of inhaled mycobacteria through tonsils and adenoids. It can also spread via lymphatic or by hematogenous dissemination from a primary focus in the lungs.<sup>1</sup> In the present case submandibular and jugulodigastric lymph nodes of the patient were enlarged.

In cases of cervical lymphadenitis, a thorough inspection is mandatory, as there are often no signs and symptoms of pulmonary TB.<sup>9</sup> The current case also had no signs of pulmonary involvement.

Diagnostic images such as CT and MRI prove to be very useful in these type of cases without systemic signs and symptoms as they help to localize and characterize the mass.<sup>10</sup>

In CT, the affected nodes have a centre of low attenuation with an enhancing rim. The node is homogenous and the calcification can be noticed.<sup>3</sup> The CT scan of the patient in this case only showed a focal bulge of periosteum with multiple enlarged lymph nodes as mentioned previously.

In MRI, the non-enhancing areas indicate caseation or liquefaction necrosis, and the enhancing areas indicate granulation tissue with an inflammatory hyper vascularity and increased vascular permeability.<sup>3</sup> MRI of the current patient revealed a well-defined thick walled subcutaneous abscess on the left submandibular region and minimal cortical bone irregularity in the inner surface of the left angle of mandible.

Chest X-ray is also considered in the diagnosis of tuberculous lymphadenitis. Parenchymal foci associated with lymphadenopathy (Ghon complex) along with hilar or mediastinal lymphadenopathy may occur.<sup>11</sup> In our case the patient had not taken a chest x ray and hence was advised to do so after consultation with a physician.

FNAC of tuberculous lymphadenitis generally shows caseous necrosis, degenerated inflammatory cells, epithelioid giant cell granulomas and giant cells.<sup>12</sup> FNAC performed in the present patient's case revealed a very thick purulent material consisting of dense inflammatory infiltrate mainly neutrophils along with necrosis.

Histopathologic examination is one of the most important diagnostic methods of Tuberculous lymphadenitis. Langhans giant cells, caseating necrosis, granulomatous inflammation and calcification can be seen in histopathological examination of TB.<sup>3</sup> In the current case, H&E stained tissue sections showed wide areas of caseous necrosis with surrounding chronic inflammatory cells chiefly lymphocytes, plasma cells, macrophages and occasionally scattered epitheloid cells at the periphery along with Langhans giant cells. There were also areas showing abscess with clumps of neutrophils.

Differential diagnoses of tuberculous lymphadenitis mostly include submandibular sialadenitis and submandibular gland calcification.<sup>13</sup> In the present case we gave a provisional diagnosis of Suppurative Osteomyelitis initially considering the abscess and the grossly decayed teeth in the same area and considered antibioma and left submandibular sialadenitis as the differential diagnoses. Later based on the FNAC report mentioned previously, a diagnosis of Suppurative Lymphadenitis was suspected. Finallty after incisional biopsy, histopathological features were suggestive of Tuberculous Lymphadenitis with abscess.

Chronic inflammatory conditions like tuberculosis can pave way for malignancy development and hence TB can also be seen co existing with oral squamous cell carcinoma. The combination of direct DNA damage, apoptosis inhibition and persistence of chronic inflammation may lead to mutagenesis of progeny cells, and these effects together with angiogenesis, may lead to a microenvironment that is suitable for tumorigenesis.<sup>14</sup>

The treatment of TB in children follows the same principles as for adults. Treatment comprises of 2 phases: an intensive phase of 2 months with 3/4 drugs and a continuation phase of 4 months with 2 drugs. In severe/ complicated TB disease the treatment may be given for a longer time by prolonging the continuation phase to 7 months (instead of 4 months). The drug dosages depend on the body weight of the child and should be adjusted as weight changes during the course of treatment.<sup>15</sup> Our patient was put on the currently recommended treatment for lymph node tuberculosis, i.e 6 months of Rifampicin and Isoniazid plus Pyrazinamide for the first 2 months, given thrice weekly.

#### 4. Conclusion

Tuberculosis in the head and neck region is a crucial clinical concern, especially in developing nations and clinicians need to be aware of the various manifestations of TB for ensuring a prompt treatment. Diagnosis of TB becomes extremely challenging when systemic signs and symptoms remain absent as was in the present case. Early diagnosis of TB can not only decrease the advancement of the disease but also prevents the spread of the disease to others.

#### 5. Source of Funding

None.

### 6. Conflict of Interest

None.

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