

## Primary Tuberculosis of the submandibular gland: a case report

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### Abstract

Chronic sialadenitis is a disease of salivary glands associated with chronic infections, systemic diseases and sialolithiasis pathogenesis of the disease. Primary tuberculous submandibulitis is not a reported disease. The diagnosis was confirmed when a 45 years old lady suspicious to submandibular gland tumor was operated on for excisional biopsy. Histological examination of the specimen by expert pathologist ruled out malignancy, granuloma or caseous necrosis. Cultures of discharge and tissue were negative for mycobacterium tuberculosis. The only positive diagnostic modality was Polymerase Chain Reaction for mycobacterium tuberculosis. Therefore, the four drug anti-TB regimen was initiated. After that a surgical fistula was healed and the patient left the hospital on her foot and there was no recurrence at least for one year. The endemic condition of tuberculosis in some populations has increased the rate of extra-pulmonary tuberculosis. One of the extra pulmonary sites may be the submandibular gland.

It is recommended to consider tuberculosis in the differential diagnosis of submandibular gland masses especially in populations with endemic tuberculosis.

**Keywords:** chronic sialadenitis, tuberculosis, submandibular gland.

### Introduction

Chronic infection of the salivary glands is rare and usually occurs as a result of recurrent infections following trauma, stone, stasis, inflammation, autoimmune disease, sarcoidosis, stricture, neoplasm and miliary tuberculosis. Submandibular gland might be infected by bacteria and become purulent. This infection mostly occurs in patients with malnutrition, dentures and systemic disease. The most common microorganisms involved in chronic purulent submandibulitis are staphylococcus and mixed aerobic and anaerobic bacteria [1]. Clinical mani-

festation of chronic sialadenitis includes non-tender swelling of the gland.

Solitary tuberculosis (TB) of the salivary glands is rarely reported and involvement of submandibular gland is too rare. In infections such as TB and actinomycosis, there could be fistula and drainage of pus. However, it is notable that clinical presentations are not sufficient clues to differentiate various diseases of the salivary glands [2]. Also radiological evaluations such as sonography, sialography, CT or MRI cannot accurately help to the differential diagnosis [3]. Since most of the submandibular gland tumors are malignant, a benign solitary enlargement of the gland should be differentiat-

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ed from cancer by excisional biopsy [4]. Although needle biopsy is safer and less invasive, and may be performed easily, it is not satisfactory [5]. In populations where TB is common, salivary glands could also be involved, sometimes appearing as primary tuberculosis of salivary gland [6]. Before the spread of AIDS, TB rarely caused extra pulmonary infections. However, with the spread of AIDS, extra pulmonary TB increased [7]. Tuberculosis results in a range of clinical disorders and is one of the leading causes of mortality and morbidity in the world.

### Case presentation

A 45-year-old otherwise healthy woman admitted in hospital in Sabzevar, Khorasan, Iran, was presented to Baqiyatallah Hospital in Tehran for a second opinion regarding a complicated history of globus sensation in her right submandibular space, with mild pain but fever for two months. She was intermittently treated with antibiotics but the submandibular space gradually became tender and a cellulitis with intact skin was formed. Her medical work-up included testing for autoimmune diseases, sarcoidosis, sjogrens and Wegener's granulomatosis which were entirely negative. The patient had a negative chest Xray and TB skin test. MRI with Gadolinium showed enhancing mass in right submandibular area resembling malignant tumor. FNA was not conclusive. At this time the submandibular gland was resected in the operating room and tissue samples from the surgery were sent for histopathological evaluation and mycobacterial and routine cultures. A complicated incisional fistula became persistent for few weeks. Histopathology of the gland demonstrated diffuse inflammation, multinucleated giant cells similar to Langerhans' cells, and macrophages, accompanying with malignancy, caseating and non-caseating granulomas or sulfur granules. Smear and cultures were sterile and there was no growth of any pathogenic microorganism after 14 days. Suspecting other

diseases the patient underwent further evaluations. CT-scan and gallium scan of lung were performed and both were normal. Considering the complaint of chronic cough, bronchoscopy was performed and showed diffuse inflammation in bronchial mucosa. Broncho Alveolar lavage (BAL) was negative for TB and other usual microorganisms. CT scan of paranasal sinuses was normal. The Polymerase Chain Reaction (PCR) of the surgical sample was compatible with mycobacterium tuberculosis.

With the final confirmation of primary tuberculosis of the submandibular gland, the patient was put on four drug anti-TB regimen, consisting of rifampin 600 mg/d, isoniazid 300 mg/d, pyrazinamide 1500 mg/d, and ethambutol 800 mg/d for a 6-month-duration. With this treatment the patient completely recovered, the surgical fistula was healed, the patient left the hospital one month after operation and there was no report of recurrence of disease at least for one year.

### Discussion

Chronic sialadenitis is a rare disorder of the submandibular gland for which the patient receives different antibiotic regimens, has frequent visits to the clinics and may undergo surgical drainage [1]. Various causes have been mentioned for this condition. Based on patient's clinical history and examination, various laboratory and pathological investigations must be performed in order to reach a final diagnosis. Needle biopsy can be used in many instances and may not be needed for the total removal of the gland. Malignancies are common in submandibular gland and this should be considered in the differential diagnosis of submandibular gland hypertrophy. In our patient, cytological examination of the specimen was negative for malignant cells. Sarcoidosis and autoimmune disorders should be considered in young individuals. These conditions were ruled out in our case. Tuberculosis (TB) remains a major health concern. After a resurgence in the

1980s, there has been a steady decline of the disease in the United States [4]. Approximately 85% of reported cases involve the lungs and the remainder involve extrapulmonary sites. Tuberculous sialadenitis may occur both as primary and concurrently with pulmonary TB. In our patient, it occurred as primary in the absence of pulmonary TB. Although the condition is absolutely rare, primary tuberculous submandibulitis should be considered in areas where TB is endemic. Furthermore, in areas where AIDS is highly prevalent, primary tuberculous sialadenitis should be considered [6].

Due to the high incidence of TB in some endemic areas, a large number of people have positive tuberculin skin test, therefore, this test, clinical examination and imaging analysis may be insufficient for differentiating tuberculous sialadenitis from other conditions. Histopathological examination of the specimen is essential for differentiating it from malignancy. Observing caseous necrosis and granuloma in the histological examination of the biopsied material is helpful for diagnosis [8,9,10]. PCR and tissue culture are another methods by which tuberculosis would be confirmed [11]. PCR which was performed in this study, was the only positive diagnostic tool and therefore, necessary to perform PCR in the suspicious cases. Cultures of tissue and discharge were negative, this response could be explained by the lack of specific culture media and/or early reading of the result. It is essential to keep the tissue culture for a longer period of time in suspicious cases of TB [12].

The disease was confirmed in our patient by pathological clues compatible with TB, positive PCR, good therapeutic response to anti-TB medications and absence of recurrence.

### Conclusion

Regarding the mentioned case which was diagnosed as tuberculous submandibulitis, it is recommended to perform TB work-up for patients who do not show therapeutic response to

sufficient anti-bacterial therapies especially in TB and AIDS endemic populations.

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