Primary Tuberculosis of Submandibular Salivary Gland

Leo Francis Tauro, Celine George, Aroon Kamath, GK Swethadri[1], Rohan Gatty

Departments of General Surgery and [1]Pathology, Fr. Muller Medical College Hospital, Kankanady, Mangalore (D.K.), Karnataka, India

ABSTRACT

Tuberculosis of the submandibular salivary gland is a rare condition and only a few cases have been reported in literature. Tubercular sialadenitis is most frequently seen in immunosuppressed patients. Diagnosis of this disease is difficult. Although fine needle aspiration cytology is useful in diagnosis, excisional biopsy is often required. Polymerase chain reaction for mycobacterium tuberculosis is a reliable diagnostic tool, and if available, it should be performed before surgical intervention to enable differential diagnosis of a salivary gland tumor. We report two cases of the submandibular salivary gland tuberculosis from South India (Mangalore located in the coastal belt of Karnataka) that proved diagnostically challenging. Both patients responded well to antitubercular therapy and surgery was avoided.

Key words: Mycobacterium, Salivary glands, Sialadenitis, Submandibular gland

INTRODUCTION

Tuberculosis of the submandibular salivary gland is a rare clinical entity even in countries where the disease is rampant. Only a few cases of submandibular salivary gland tuberculosis have been reported in literature. In developing countries, tuberculous infection still exists on a significant scale. There is a slight reduction of incidence of tuberculosis in developed countries owing to increased awareness towards hygiene and nutrition. In majority of the cases of tuberculosis involving the salivary glands, the parotid is the commonest gland to be affected.[1] We report two cases of tuberculosis of the submandibular salivary gland from South India (Mangalore located in the coastal belt of Karnataka, where tuberculosis is common) for their clinical interest and diagnostic dilemma.

CASE REPORTS

Case 1

A 45-year-old male patient presented with a swelling in the right submandibular region (just inferior to the angle and adjoining part of the body of the mandible) associated with pain for one month. The onset of symptoms was sub acute in nature. There was no history of fever, cough, weight loss or other systemic symptoms ("B" symptoms). There were no symptoms suggestive of pulmonary tuberculosis. The patient was immunocompetent and there was no family history of tuberculosis. The patient consulted a dentist, who diagnosed it as lower jaw tooth abscess. He was treated with antibiotics and lower 2nd molar tooth extraction was done. Even after 2 weeks of treatment the swelling increased in size, hence the patient was referred to the surgical outpatient department. His general physical examination was unremarkable. Local examination detected a diffuse, non-tender, hard, bi-digitally palpable swelling of 6 cm × 5cm in the submandibular region just inferior to the angle and adjacent part of the body of the mandible on the right side. The skin overlying the swelling was normal. The swelling was indistinguishable from the mandible. There was no purulent discharge from the Wharton's duct and its opening was normal. The rest of the oral cavity and oropharyngeal examination was unremarkable. The deep cervical lymph nodes were palpable on right side (level 2); the biggest being 3 cm × 2cm in size, fixed and hard in consistency [Figure 1]. The rest of the systemic examination revealed no abnormality.

The complete blood count was normal except for a raised ESR of 45 mm. Liver function tests (LFT)

Access this article online

Quick Response Code:

Website: www.jgid.org

DOI: 10.4103/0974-777X.77301

Address for correspondence:
Dr. Leo Francis Tauro, E-mail: drlftauro@rediffmail.com

including the liver enzymes and serum proteins were within normal limits. Chest X-ray did not reveal any pulmonary lesion. Enzyme linked immuno sorbent assay (ELISA) for human immunodeficiency virus (HIV) and hepatitis B surface antigen (HBs Ag) were non-reactive. Fine needle aspiration cytology (FNAC) from the swelling showed benign ductal cells, stromal fragments with fibrosis, myxoid ground substance with few neutrophilic and lymphocytic inflammatory cells suggestive of chronic sialadinitis. FNAC from the lymph node showed reactive population of lymphoid cells, suggestive of reactive hyperplasia; few histiocytes, and vague epithelioid granulomas. The pathologist suggested incisional biopsy to rule out tuberculosis. The radiograph did not show any abnormality in the mandible. CT scan of head and neck showed enlarged right submandibular salivary gland with diffuse fat stranding. Inflammatory hypo dense mass was noted surrounding the gland with thickening of overlying subcutaneous tissue and skin. Enlarged right submandibular lymph node and a necrotic right deep cervical lymph node (level 2), 1.2 cm size were noted. Fat planes of mylohyoid muscles were lost. Bony structures of neck showed no abnormality. These findings suggested the possibility of inflammatory etiology–right submandibular sialadinitis [Figure 2]. Incisional biopsy from the swelling showed chronic inflammatory process with a few vague granulomas comprising epithelioid cells, Langhan's giant cells, and lymphocytes. Special stains for acid fast *bacillus* (AFB) were negative. These features were suggestive of chronic granulomatous disease, most likely tuberculosis [Figure 3a and b]. Serology for toxoplasmosis proved negative. Polymerase chain reaction (PCR) for mycobacterium tuberculosis was positive. Based on these findings, tuberculous sialadenitis was diagnosed and antitubercular treatment (ATT) was started, i.e., Isoniacid (INH) 300 mg, Rifampicin 450 mg, Pyrazinamide 1500 mg, and Ethambutol 800 mg for 2 months; INH 300 mg and Rifampicin 450 mg for 7months. Patient responded well to seven months' ATT and the swelling completely disappeared [Figure 4]. At 6 months post-ATT follow-up, the patient was asymptomatic without any swelling.

Figure 1: Photograph of the patient showing right submandibular swelling (a) Frontal view. (b) Right lateral view

Case 2

A 25-year-old male patient presented with a swelling in the left submandibular region (just inferior to the angle and body of the mandible) associated with pain for two months. There were no "B symptoms" (constitutional symptoms) and symptoms suggestive of pulmonary tuberculosis. The patient was immunocompetent and there was no family history of tuberculosis. The patient consulted a dentist and he was treated with antibiotics. Due to continuing pain and increase in size of the swelling, the patient was referred to surgical outpatient department. His general physical examination was unremarkable. Local examination

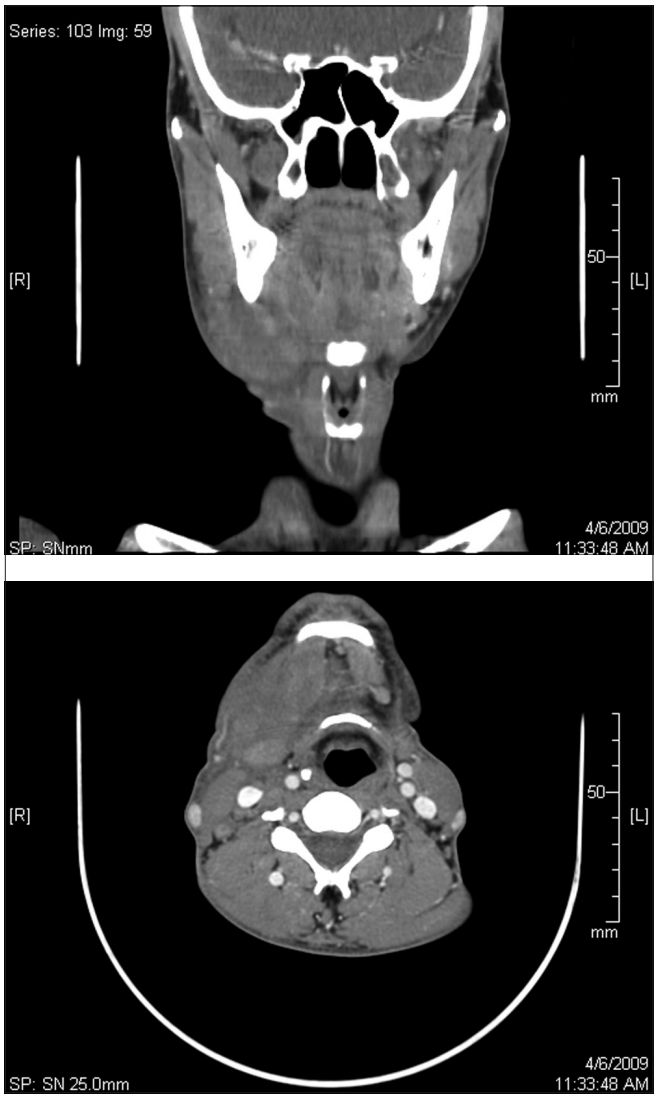

Figure 2: CT scan showing right submandibular gland inflammatory swelling

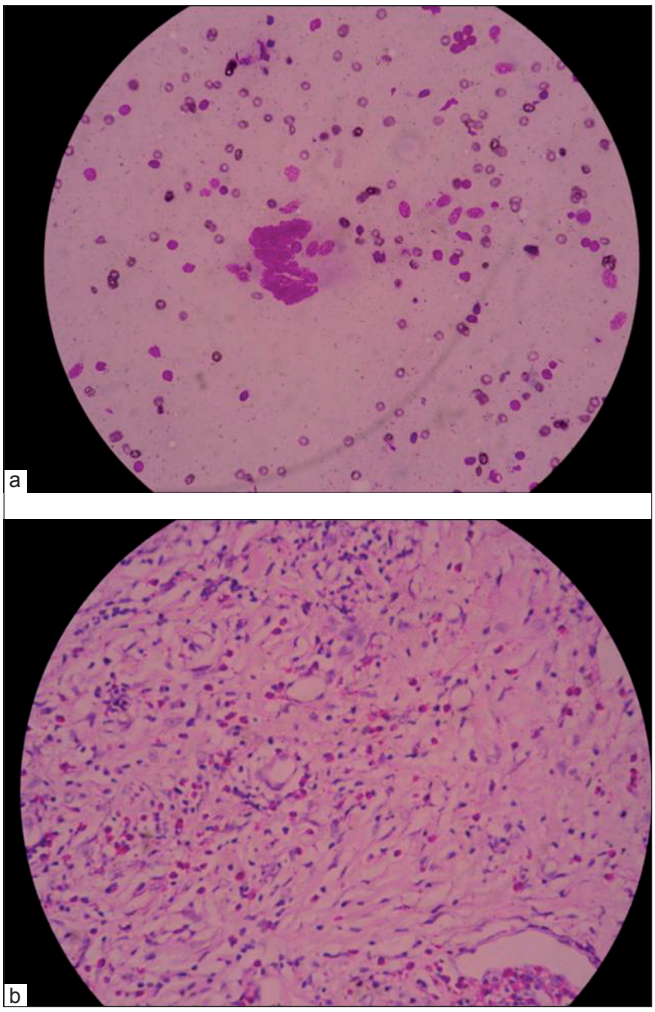

Figure 3: Photomicrograph (a) showing granuloma. (May Grunwald Giemsa stain 450×). (b) Chronic sialadenitis (Hematoxylin – Eosin stain 450×)

detected a diffuse, non-tender, firm, bi-digitally palpable swelling of 8 cm × 5cm in the left submandibular region. The skin overlying the swelling was normal. The swelling was not fluctuant. There was no purulent discharge from the Wharton's duct. The rest of the oral cavity and oropharyngeal examination was unremarkable. The cervical lymph nodes were not palpable. The rest of the systemic examination revealed no abnormality.

The complete blood count was normal except for a raised ESR of 68 mm. LFT including the liver enzymes and serum proteins were within normal limits. ELISA for HIV and HBs Ag were non-reactive. The FNAC from the swelling showed a few granulomas comprising epithelioid cells, Langhan's giant cells, and lymphocytes with central caseation necrosis. Special stains for AFB were negative. These features were suggestive of tuberculous sialadinitis. Chest X-ray did not reveal any pulmonary lesion. The radiograph did not show any abnormality in the mandible.

Figure 4: Photograph of the patient after treatment showing complete regression of swelling

PCR for mycobacterium tuberculosis was positive. Patient was started on ATT, i.e., INH 300 mg, Rifampicin 450 mg, Pyrazinamide 1500 mg, and Ethambutol 800 mg for 2 months; INH 300 mg and Rifampicin 450 mg for 7months. The swelling had completely disappeared at the end of the treatment period. At 8 months post-ATT follow-up, the patient was asymptomatic without any swelling.

DISCUSSION

Extra pulmonary tuberculosis is relatively common cause of granulomatous disease of the head and neck. It is relatively rare in the salivary glands, even in those areas where tuberculosis is endemic.[2] Tuberculosis of salivary gland must be included in the differential diagnosis of a submandibular gland mass, especially when the patient is from a low socioeconomic group with poor hygiene, malnourished, and from an endemic area.[2] The salivary gland tuberculosis is much more common in immunosuppressed patients. Usually tuberculosis affects one side, and the parotid gland is the most common salivary gland to be infected with tuberculosis.

Involvement of the salivary gland is thought to arise from a preceding tooth or tonsil infection. Direct extension to the salivary gland parenchyma by the *bacillus* may occur through the duct system. Primary tuberculosis of salivary gland may occur in two forms as an acute inflammatory lesion (mimicking acute suppurative sialadinitis) or as a chronic mass (tumor) lesion that may be asymptomatic for many years.[2] The tuberculous disease is impossible to distinguish clinically from other diffuse diseases of salivary glands if glandular secretions from Wharton's duct or saliva are negative for AFB.[2,3] Most forms of sialadenitis are resolved by a surgical procedure. However, a cure for

tuberculosis of submandibular salivary gland requires adequate and prolonged ATT.[2-4]

Tuberculosis of the submandibular salivary gland is a rare pathology and does not always have the diagnostic guideline led by previous tubercular localizations.[3] Diagnosis of tuberculous sialadenitis may be difficult. Clinically salivary gland tuberculosis is indistinguishable from a neoplasm of the gland, although benign neoplasms of the gland tend to grow slowly over years rather than over a few months, as in case of tuberculous involment. A malignant salivary gland tumor may be fast growing, unlike tuberculosis. Mycobacterium tuberculosis infection within parotid gland Warthin tumor is known to co-exist.[5] Constitutional symptoms are usually absent, and often there is no evidence of active tuberculosis elsewhere in the body.[2,3] A presumptive diagnosis of tuberculous sialadenitis can be made if there is evidence of tuberculosis elsewhere in the body.[2] But in cases where no lesion is detectable elsewhere, the diagnosis most often is made by culture of saliva, tissue culture, aspiration cytology, or histopathology. The material can be subjected to AFB staining. Incisional biopsy should be avoided as it can lead to a chronic fistula. If the lesion needs to be opened, excisional biopsy should be carried out.[2-4] Chintamani *et al*.,[6] have reported three cases of parotid tuberculosis of which two were diagnosed by FNAC and third case was diagnosed by histopathology only after surgery. PCR for mycobacterium tuberculosis is a reliable diagnostic tool, and if available, it should be performed before surgical intervention to enable differential diagnosis of a salivary gland tumor.[7]

Non-tubercular (atypical) mycobacterial infections in children commonly present as cervical masses.[8] In these cases, skin testing (purified protein derivative (PPD)) with new tuberculins and culture should be performed to help the diagnosis. Prasad *et al*.,[9] have reported the increased incidence of isolated head and neck tuberculosis. In their study, 73.3% patients had isolated tuberculous lymphadenitis, 1.8% (3 cases) had parotid tuberculosis, and 75% of head and neck tuberculosis patients did not have pulmonary involvement. FNAC was highly effective in the diagnosis of nodal tuberculosis, but histopathological examination was required to make the diagnosis in other head and neck sites. The PPD test was not effective as diagnostic tool. It was positive in 20% of the cases.

In cases diagnosed as tubercular sialadenitis, majority resolve on ATT and surgery should be reserved for the lesions showing poor or no response to ATT.[5-7,10] In cases of atypical mycobacterial infections, surgical excision and curettage may be adequate owing to a less virulent nature of the pathogens.[8] Treatment involves administration of ATT (i.e., combination of INH, Rifampicin, Pyrazinamide, and Ethambutol) for 8–12 months.[7-9] If resistant to medical treatment the gland or tumor lesion should be excised.[4,7-10]

CONCLUSION

To conclude, the cases under discussion presented with more of a sub-acute response. The source of infection in these cases was uncertain. However, it was probably from tooth or tonsils or elsewhere in the oral cavity. Since our patients had no evidence of concomitant tuberculosis elsewhere in the body and tuberculosis of submandibular gland is a rare disease, we did not consider it in our provisional diagnosis. In the first case, the swelling was hard and adherent to the mandible and FNAC was inconclusive, therefore an incisional biopsy was performed. In both cases, PCR for mycobacterium tuberculosis was positive. PCR for mycobacterium tuberculosis is a reliable diagnostic tool, and if available, it should be performed before surgical intervention to enable differential diagnosis of a salivary gland tumor. We have reported these cases for its clinical interest and diagnostic dilemma. Both cases had responded well to antitubercular treatment without surgery.

REFERENCES

1. Kumar S, Dev A. Primary tuberculosis of bilateral submandibular salivary Glands. Indian J Otolaryngol 1990;42:68-9.
2. Ataman M, Sözeri B, Ozçelik T, Gedikoglu G. Tuberculosis of the parotid salivary gland. Auris Nasus Larynx 1992;19:271-3.
3. Bottini DJ, Garelli A, Felici M, Galante V, Cervelli V. Primary tuberculosis of the submandibular salivary gland. J Craniofac Surg 2007;18:218-9.
4. Tsutomu S, Kyoko N, Nobuhiko F. Tuberculous sialadenitis of submandibular Gland. Kita Kanto Med J 1999;49:177-80.
5. Ozcan C, Apa DD, Aslam G, Gulhan S, Gorur K. Mycobacterium tuberculosis infection within parotid gland Warthin tumor. J Craniofac Surg 2008;19:1561-5.
6. Chantamani, Daniel R, Manu S, Bhushan V, Gupta K. Parotid TB. Trop Doct 2006;36:119-20.
7. Kim YH, Jeong WJ, Jung KY, Sung MW, Kim KH, Kim CS. Diagnosis of major salivary gland tuberculosis: Experience of eight cases and review of literature. Acta Otolaryngol 2005;125:1318-22.
8. Jervis PN, Lee JA, Bull PD. Management of non-tuberculous mycobacterial peri-sialadenitis in children: The Sheffield otolaryngology experience. Clin Otolaryngol Allied Sci 2001;26:243-8.
9. Prasad KC, Sreedharan S, Chakravarthy Y, Prasad SC. Tuberculosis in the head and neck: Experience in India. J Laryngol Otol 2007;121:979-85.
10. Moure C, Mbuyamba S, Bruniau A, Gbaguidi C, Testelin S, Boutemy M, *et al*. Tuberculosis of the submandibular Gland: A case report. Rev Stomatol Chir Maxillofac 2006;107:115-8.

How to cite this article: Tauro LF, George C, Kamath A, Swethadri GK, Gatty R. Primary tuberculosis of submandibular salivary gland. J Global Infect Dis 2011;3:82-5.

Source of Support: Nil. **Conflict of Interest:** None declared.