



SUBMANDIBULAR GLAND TUBERCULOSIS WITH LYMPH NODE METASTASIS: A RARE CASE REPORT

Pathology

**Swati Bipin
Ghanghurde**

Senior Resident, ESI-PGIMSR, Andheri (E), Mumbai

**Sushma Rajesh
Gulhane**

Specialist, ESI-PGIMSR, Andheri (E), Mumbai

Madhuri Kate*

Professor & Head of the Department, ESI-PGIMSR, Andheri (E), Mumbai

*Corresponding Author

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 without a high index of suspicion is difficult. Although fine needle aspiration cytology is useful in diagnosis, 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 a case of the submandibular salivary gland tuberculosis with lymph node metastasis for its rarity and diagnostic dilemma.

KEYWORDS

Mycobacterium, Salivary glands, Sialadenitis, Submandibular gland

Introduction

Tuberculosis is a necrotizing granulomatous disease with varied clinical presentations and a wide distribution. Tuberculosis is common in countries like India, the lungs are most commonly involved. Extra thoracic forms of the disease account for approximately 20% of overall active tuberculosis and can be seen in the kidneys, bones, meninges, and lymph nodes, submandibular gland is uncommon.¹ Tubercular sialadenitis is most frequently seen in immunosuppressed patients. We report a rare case of submandibular gland tuberculosis with lymph node metastasis.

Case report:

A 48-year-old otherwise healthy woman admitted with left submandibular gland lump since 2 months (fig 1).



Fig 1 left submandibular gland lump

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. Her general physical examination was unremarkable. Local examination detected a diffuse, non-tender, firm, bi-digitally palpable swelling of 6 cm X 5cm in the submandibular region just inferior to the angle and adjacent part of the body of the mandible on the left side. The skin overlying the swelling was normal. The swelling was indistinguishable from the mandible. There was no purulent discharge. The rest of the oral cavity and oropharyngeal examination was unremarkable. The deep cervical lymph nodes were palpable on left side the biggest being 3 cm × 2cm in size, fixed and firm in consistency. The rest of the systemic examination revealed no abnormality. She was intermittently treated with antibiotics but the lump has not reduced. USG suggestive of sialolithiasis of submandibular duct with sialoadenitis with abscess formation. 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 sialadenitis. FNAC from the lymph node shows acute on chronic non-specific inflammation. At this time the left submandibular gland along with left cervical lymph node was resected and sent for histopathological evaluation. Histopathology of the gland demonstrated replacement of gland with well formed granulomas with central caseous necrosis, epithelioid cells, giant cells, surrounded by lymphocytes (fig 2).

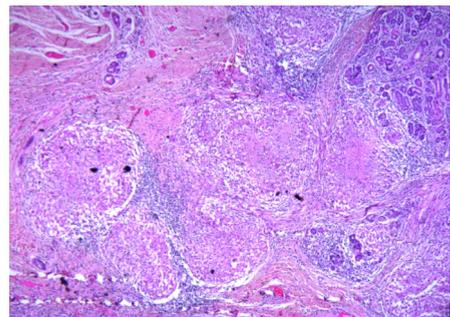


Fig 2 submandibular gland showing well formed granulomas admixed with salivary gland

LN also shows similar morphology (fig 3).

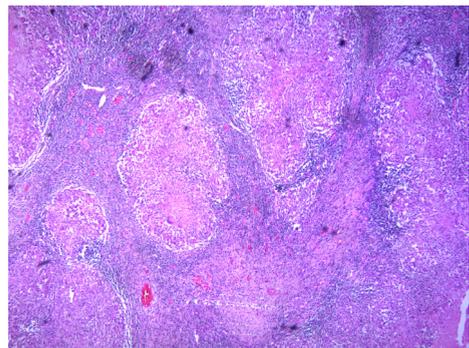


Fig 3. lymph node showing granulomas

ZN stain was negative for acid fast bacilli. This features were suggestive of chronic sialoadenitis with granulomatous lymphadenitis suggestive of tuberculosis. PCR was not done in this case. The patient was started on first line antitubercular drugs with four months of follow up.

Discussion:

There are a variety of non-neoplastic disorders affecting the salivary glands. These include chronic sialoadenitis, tuberculosis, sarcoidosis, animal scratch disease, actinomycosis and Sjogren's syndrome (I) and they require differential diagnosis and management. The salivary gland is a rare site for primary TB, possibly due to the presence of thiocyanate ions and pro-teolytic enzymes such as lysozymes in the salivary gland secretions and continues flow of saliva which prevents lodging of the bacteria. In India, it is estimated that more than 40% of the adults are infected with tuberculosis bacilli and about 2 million people develop tuberculosis every year and about 500,000 die from it.² Although only around 100 cases were reported of salivary gland tuberculosis worldwide and majority of the cases involve the parotid gland till date, the submandibular gland involvement includes only less than 10 cases.³ First is the 48 year old female with left submandibular gland tuberculosis reported by Sakurai et al. in 1999 in Japan.³ It is relatively rare in the salivary gland, even in those areas where tuberculosis is endemic.⁴ It is relatively rare in the salivary gland, even in those areas where tuberculosis is endemic.⁴ 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 areas.⁴ The salivary gland tuberculosis is much more common in immunosuppressed patients. Involvement of the salivary glands is thought to arise from a preceding tooth or tonsil infection. Primary TB of the salivary gland may occur in two forms: as an acute inflammatory lesion mimicking acute suppurative sialoadenitis, or as a chronic tumorous lesion.

Diagnosis of salivary gland tuberculosis requires a very high index of suspicion and could be a dilemma due to non-specific wide range of presentation and its rarity. Constitutional symptoms are usually absent, and often there is no evidence of active tuberculosis elsewhere in the body.⁴ Patra et al reported a case of salivary gland tb with dental pain. Dental extraction and antibiotic did not improve the condition. CT scan and FNAC were indicative of submandibular abscess.⁵ A presumptive diagnosis of tuberculous sialadenitis can be made if there is evidence of tuberculosis elsewhere in the body.⁴ 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 constitutional symptoms are frequently absent as in our case. Differential diagnosis includes sialadenitis, submandibular lymphadenitis, dental abscess and submandibular gland tumor.

FNAC is a simple procedure and when combined with PCR will be of great value. The sensitivity of FNAC alone for diagnosing tuberculosis is known to be less than 50% however it can reach to 90% sensitivity and specificity if combined with PCR.³ In our case FNAC was performed but was not combined with PCR because of low index of suspicion so the result was not conclusive and further surgical resection of the gland was needed for tissue confirmation. Even though CT scan is a very useful tool to investigate the lesion unfortunately it is extremely difficult to differentiate mycobacterial inflammatory lesions from neoplastic masses by means of CT alone.³

CONCLUSION

To conclude, our case had sub-acute response and the source of infection in these cases was uncertain. However, it was likely from tooth or tonsils or elsewhere in the oral cavity. Since our patients had no evidence of associated tuberculosis elsewhere in the body and tuberculosis of submandibular gland is a rare disease, we did not consider it in our provisional diagnosis. So we report this case for its rarity, and to have differential of submandibular gland tuberculosis in endemic areas when there is diagnostic dilemma.

REFERENCES

1. Gupta, V., Patankar, K., Shinde, A., Bhosale, C. and Tamhane, A. (2012). Tuberculosis of the Parotid Gland. *Case Reports in Radiology*, 2012, pp.1-3.
2. Dadwal, M. (2011). Primary Submandibular Tuberculosis: An Unusual Cause of Submandibular Salivary Gland Enlargement. *Indian Journal of Otolaryngology and Head & Neck Surgery*, 63(3), pp.298-299.
3. Kasim, K., Namavar, E. and Mohamad Yunus, M. (2015). A rare case of primary tuberculosis of the submandibular gland!!!. *Egyptian Journal of Ear, Nose, Throat and Allied Sciences*, 16(1), pp.87-89.
4. Tauro, L., George, C., Kamath, A., Swethadri, G. and Gatty, R. (2011). Primary tuberculosis of submandibular salivary gland. *Journal of Global Infectious Diseases*, 3(1), p.82.
5. Patra, S. (2013). Primary Tuberculosis of Submandibular Gland Presenting as Dental Abscess. *Journal of Otolaryngology & Rhinology*, 02(02).