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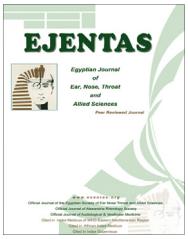

CASE REPORT

A rare case of primary tuberculosis of the submandibular gland!!!

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KEYWORDS

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Submandibular gland;
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Abstract A rare case of primary tuberculosis of the submandibular gland is reported here which required surgical gland excision for definitive diagnosis. It is presented in view of its rarity, the extreme difficulty in the diagnosis of this kind of disease and highlighting the importance of histopathological examination.

1. Introduction

Salivary gland tuberculosis is a rare condition and majority of the reported cases involve the parotid gland. Primary submandibular TB is rare in English literature and less than 10 cases have been reported. Diagnosis of submandibular TB is very challenging and requires high index of suspicion as the symptoms are non-specific. TB salivary gland can be cured medically and early diagnosis is of importance to avoid unnecessary surgical intervention and ensure complete remission. We report a case of tuberculosis of the submandibular salivary gland for its clinical interest and diagnostic dilemma.

2. Case report

36 year old gentleman presented to our ENT clinic with a left submandibular region swelling, gradually increasing in size associated with occasional pain and low grade fever for 1 month duration. He was treated by GPs as sialadenitis and received multiple courses of antibiotics with no improvement. There were no constitutional symptoms, night sweat and no TB contact.

On examination a firm, mildly tender, defuse, bi-manually palpable swelling measuring 3×3 cm in the left submandibular region was observed. The skin overlying the swelling was normal. The swelling was adherent to 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. There was no palpable cervical lymphadenopathy and other ENT examinations were unremarkable. Other blood investigations were normal. He was immunocompetent. Chest X-ray showed no abnormalities. Contrasted CT scan (Fig. 1) of the neck revealed 3×4 cm swelling within the left submandibular gland with inhomogeneous consistency suggestive of tumor. Fine needle aspiration cytology (FNAC) from the swelling revealed evidence of fibrosis and few neutrophilic and lymphocytic inflammatory cell infiltration which was suggestive of chronic sialadenitis.

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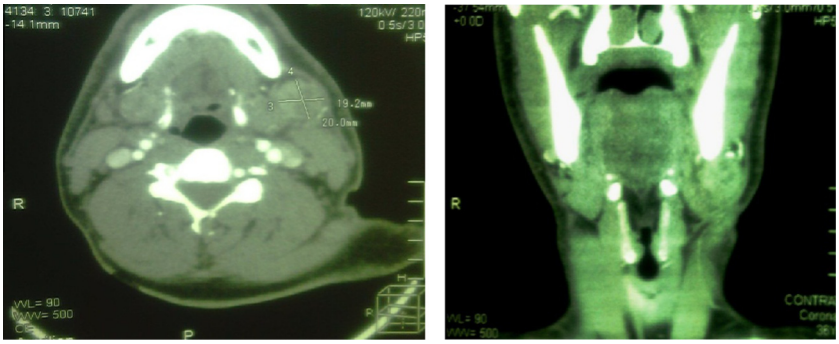

Figure 1 Contrasted axial section (right) and coronal section (left) at the level of the submandibular gland.

Taking the above into consideration, patient who underwent uneventful left submandibular gland excision and specimen (Fig. 2) was sent for histopathological examination (HPE) which surprisingly revealed a granulomatous disease of the left submandibular gland (Fig. 3). TB culture was positive from the saliva. The patient was on anti-tubercular therapy for 6 months. He completed the course successfully and uneventfully. In 1 year follow up showed complete recovery with no evidence of remission.

3. Discussion

Tuberculosis plays a major role in morbidity and mortality in developing countries. Head and neck tuberculosis is a common extra pulmonary infection and represents 30% of all mycobacterial infections.[1] The salivary gland is a rare site for primary TB, possibly due to the presence of thiocyanate ions and proteolytic enzymes such as lysozymes in the salivary gland secretions and continues flow of saliva which prevents lodging of the bacteria. Although only around 100 cases were reported of salivary gland tuberculosis worldwide and majority of the cases involve the parotid gland[2] 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.[4]

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. The wide range of presentation can be as an acute inflammatory lesion as suppurative sialadenitis or a chronic asymptomatic mass.[5] The constitutional symptoms are frequently absent as in our case. Differential diagnosis includes sialadenitis, submandibular lymphadenitis, dental abscess and submandibular gland tumor. To diagnose this kind of disease which is extremely difficult histologic examination is mandatory, as happened in our case. Incisional biopsy must be avoided as it can lead to chronic fistula. If the lesion needs to be opened, excisional biopsy should be carried out.[6] 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.[5,6] 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.[6]

As salivary gland tuberculosis is medically curable. Surgical resection of the gland is not indicated unless dealing with tumorous conditions and diagnosis in not established. Non-typical mycobacterium infection is another indication

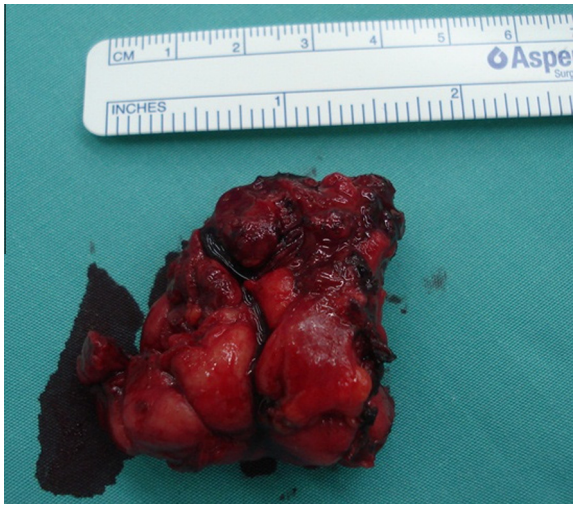

Figure 2 Gross specimen of the left submandibular gland post excision.

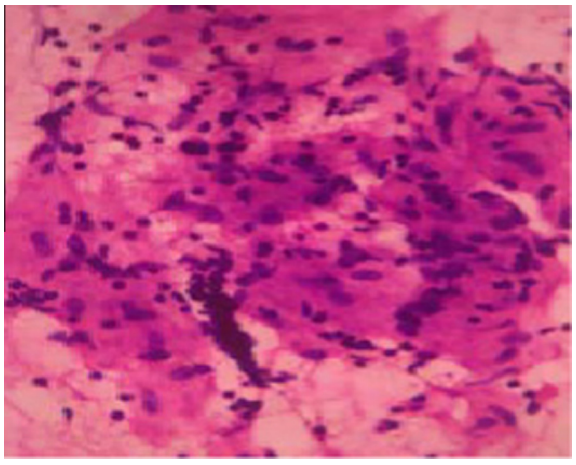

Figure 3 Hematoxylin and eosin stain X60 magnification showing epithelioid cell granuloma with central necrosis on a salivary gland tissue background.

for surgical removal of the gland.[5–7] This further emphasizes the importance of PCR investigation to manage the patient accordingly. The aim of this case is to make the readers aware about the wide range of presentation of submandibular gland tuberculosis. The physicians should consider tuberculosis as a differential in any doubtful case of submandibular swelling and it is recommended to consider PCR to avoid un-necessary surgeries or complications.

4. Conclusion

Given the worldwide reappearance of tuberculosis and especially in areas with endemic infections salivary gland tuberculosis has to be included in the differential diagnosis. With clinical suspicious tuberculosis FNAC combined with PCR could be of great value to avoid unnecessary surgical intervention. If diagnosis is doubtful surgical resection and tissue investigation is a standard procedure.

Conflict of interest

None.

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