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CASE REPORT

# Bilateral parotid gland tuberculosis: A rare occurrence



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**KEYWORDS**

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**Abstract** Parotid gland tuberculosis is an extremely rare form of extrapulmonary tuberculosis, even in countries where tuberculosis is endemic like India; however, it should be included as one of the differentials of discrete parotid swelling as it generally presents as a slow-growing mass indistinguishable from a malignancy and even imaging too, can't differentiate these clearly. The majority of the previously reported cases were mostly unilateral and diagnosed by histopathological examination of post parotidectomy specimens. Here we are describing a case of tuberculosis of both parotid glands in a 25 year-old male who was referred to us with bilateral parotid region swelling of two month duration. Tubercular parotiditis was confirmed by demonstration of epithelioid granuloma and caseous necrosis compatible with TB on fine needle aspiration cytology (FNAC). He was treated with four drug anti-TB regimen (2HERZ + 4HR) leading to full recovery and complete disappearance of swelling and symptoms with no recurrence till one year of follow up. Apart from rarity due to bilateral involvement, this case report highlights the clinical presentation, ultrasonography and other imaging findings, and significance of FNAC in diagnosis of this uncommon entity reinforcing the fact that the diagnosis of parotid gland tuberculosis requires a high degree of clinical suspicion.

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**Introduction**

Extra pulmonary tuberculosis (EPTB) is an emerging problem as they have a diagnostic dilemma and management controversy. In India, the incidence of EPTB ranges from 20% to

30% [1]. Among EPTB, the involvement of the parotid gland is quite rare even in an endemic country like India and merely hundred cases have been reported in known literature, most of them diagnosed from post parotidectomy specimens [2]. Chronic parotiditis is a very rare disease of the parotid glands, for which various causes have been enumerated, including decreased flow of secretion in the duct (due to inflammation), gland emphysema (due to duct destruction), infectious (e.g. tuberculosis), and non-infectious causes (e.g. Sarcoidosis, autoimmune diseases, Sjogren's syndrome, malignancy and duct stones). Most of previously reported cases had unilateral tubercular parotiditis. Here we are discussing a case of

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bilateral tubercular parotiditis in a 25 year old male diagnosed by fine needle aspiration cytology and successfully treated with anti-TB drugs.

### Case presentation

A 25 years old non-smoker, non-alcoholic male having symptoms of swelling in bilateral parotid region since two months referred from the ENT department with suspicion of chronic parotiditis to the outpatient department of pulmonary medicine for assessment. Initially the swelling was non tender which later on became pain full. Before attending our OPD, he had received broad spectrum antibiotics for 15 days but didn't get any improvement. There was no history of constitutional symptom, hoarseness of voice and difficulty in deglutination. He also denied for personal or contact history of tuberculosis and any oro-dental surgical procedure recently.

Local examination of parotid region revealed two firm, non-mobile, tender swellings of  $3 \times 3$  cm and  $2 \times 2$  cm on the right side (Fig. 1) and  $1 \times 1$  cm on the left side. Swelling extended antero-posteriorly from sub-mandibular angle to ear lobe on the right side and just beneath the angle of mandible on the left side. There was no sinus or discharge from swelling and facial nerve was intact. Oral examination showed bilateral diffuse parotid enlargement which was soft to firm in consistency with normal overlying skin.

Through physical examination excluded clubbing, cyanosis, dilated veins, pedal oedema, organomegaly and lymphadenopathy. Examinations of other systems were normal including respiratory system with normal chest skiagram PA view. Laboratory investigations revealed no abnormality except raised ESR (140 mm in first hour). Routine blood tests, including sugar (fasting), renal and liver function tests, urine analysis, ECG and Ultra-sonography (USG) of the abdomen showed no abnormality. HIV serology was also negative. Montoux test was positive with induration of  $20 \times 20$  cm in 48 h. The sputum smear examination was negative for acid fast bacilli (AFB). No calcification was found in sialogram of the parotid gland. Anti-Nuclear Antibody, Rheumatoid Factor and Anti-ds DNA were negative and serum ACE titre was normal, further excluding the possibility of sarcoidosis and other autoimmune disorders.



**Figure 1** Swelling in the right parotid region with some local application over it.

USG of the parotid region showed enlarged bilateral parotid gland (right > left) with hypo echoic rounded solid mass like lesions within the architecture of the right parotid gland with the absence of calcification and cystic degeneration (Fig. 2a). An intra-parenchymal lesion seen just near to the glandular region was bulging through a breach in the gland surface into the extra glandular space (Fig. 2b). Arterial type of vascularity found in the lesion during colour doppler study (Fig. 3). USG guided fine needle aspiration cytology of the swellings from both sides showed granulomatous epithelioid cell clusters, macrophages and caseation necrosis, suggestive of tubercular parotid lymphadenitis (Fig. 4). AFB smear prepared from FNAC sample was also positive, confirming our diagnosis of tubercular parotiditis. The patient was treated with four drugs (Isoniazid, Rifampicin, Ethambutol and Pyrazinamide) anti-TB regimen (CAT I RNTCP) for 2 months, followed by two drugs (Isoniazid, Rifampicin) for 4 months. With the six month of therapy, he showed a complete resolution of swellings and no recurrence even after one year follow up.

### Discussion

Tuberculosis has been a major cause of suffering and death since times immemorial and it is one of chronic necrotising granulomatous diseases with various manifestations and a wide distribution. It affects both pulmonary and extra-pulmonary organs. Extra pulmonary sites vary from cervical lymph node, bone, skin, meninges, abdomen and many more, but the involvement of parotid glands is exceedingly rare and bilateral involvement is even more uncommon [3]. First case of tubercular parotiditis was diagnosed in 1893 by C De Poali [2].

Parotid tuberculosis can occur both as primary with the absence of pulmonary TB like in our patient or concurrently with pulmonary TB. Parotid gland and lymph nodes involvement may occur in two ways. The First pathways involve ascending infection of salivary gland through Stenson's duct or accompanied lymph node, where tubercular bacilli liberate infection to the oral cavity. Haematogenous or lymphatic spread from another distant primary lung focus is the second pathway [4]. According to some authors spread by lymphatic vessels, particularly from infected tonsils and external auditory canal, plays an important role [5]. The salivary glands tuberculosis is more commonly seen because of secondary pathways than as primary itself but in our case chest skiagram was normal excluding the pulmonary tuberculosis as the source of tubercular infection.

Clinical presentation of tubercular parotiditis mimics to the neoplastic pathology and it usually presents as a unilateral localised and progressive chronic swelling or abscess in the parotid region. Presentation may vary from an acute infectious process to an indolent chronic presentation. It is almost impossible to differentiate clinically from other inflammatory diseases of salivary gland when AFB is negative from Stenson's duct secretion or saliva. The differential diagnosis includes malignancy of parotid gland, mumps, sarcoidosis, actinomycosis, etc.

Ultrasonographic examination of the parotid swelling plays a major role in the diagnosis of parotid tuberculosis. Due to their superficial location high resolution ultrasound is able to

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