

## Tuberculous parotitis: A series of eight cases and review of literature

### ABSTRACT

Although tuberculosis is a common health problem in developing countries such as India, tuberculous parotitis (tubercular involvement of parotid gland) is rarely encountered. Because of very low incidence and nonspecific symptoms, it is often misdiagnosed as parotid neoplasm. Ultrasonographic and computed tomographic findings are also noncontributory for this entity. Hence, to increase awareness about this rare entity, here, we report a series of eight cases of tuberculous parotitis which were diagnosed on fine-needle aspiration cytology (FNAC) and successfully treated with antitubercular drugs. Majority of our cases (five cases) presented as asymptomatic unilateral swelling or acute tender painful swelling (two cases) in the parotid region. FNAC smears showed caseous necrosis, epithelioid granulomas along with variable amount of mixed inflammatory exudates, and few benign ductal or acinar cell clusters. One case had unilateral recurrent swelling in the preauricular region with fistula. Superficial parotidectomy was done and histological examination revealed the diagnosis of tubercular parotitis. Following diagnosis, all patients were kept on antitubercular treatment and responded well to treatment with no evidence of recurrence on 9-month regular follow-up.

**Keywords:** Parotid, salivary gland, tubercular

### INTRODUCTION

Tuberculosis is a chronic illness with worldwide distribution. It is more common in developing countries such as India. It is a destructive disease. It mainly involves lungs and is characterized by granulomatous inflammation with or without caseous necrosis. Extrapulmonary forms of the disease account for approximately 20% of overall active tuberculosis and can affect any organ of the body. However, involvement of the salivary glands is a very rare event.<sup>[1,2]</sup> Tuberculous parotitis clinically mimics the parotid tumor which is often misdiagnosed and cases undergo unnecessary surgery. C. De Pauli reported the first case of tubercular parotitis in 1893 and since then about 100 cases have been reported in the literature.<sup>[3-6]</sup>

For ease of discussion, all the eight patients were kept into the following three groups and their clinicopathological findings are summarized in Table 1:

- Group 1: Asymptomatic unilateral preauricular swelling: This group consisted of five patients including four

males and one female with the age range of 12–65 years, presenting with unilateral swelling in the preauricular region [Figure 1a]. The duration of illness was for 3–6 months. These swellings were gradually progressive, with no pain or fever. Chest X-ray and routine biochemical examination were normal in all except one female patient in which thyroid-stimulating hormone level was raised (10.8 IU/ml) with generalized body edema and reduced appetite. On clinical examination, parotid swellings were firm, mobile, nontender, and the overlying skin was normal. There was no involvement of facial nerve. Size varied from 2.5 to 4 cm in diameter. There

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**Received:** 03-05-2018, **Revised:** 05-05-2018, **Accepted:** 25-08-2018

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**How to cite this article:** Maurya MK, Kumar S, Singh HP, Verma A. Tuberculous parotitis: A series of eight cases and review of literature. *Natl J Maxillofac Surg* 2019;10:118-22.

Access this article online	
<b>Website:</b> www.njms.in	<b>Quick Response Code</b> 
<b>DOI:</b> 10.4103/njms.NJMS_34_18	

was no evidence of active pulmonary tuberculosis in any case. In a 54-year-old female [Figure 2a], computed tomography (CT) showed enlarged parotid gland having round-to-oval soft-tissue attenuation lesion with hypodense area measuring 2.0 cm × 2.5 cm × 3.0 cm suggestive of necrosis was noted in the deep lobe of the right parotid gland. There was also mild enlargement of right submandibular gland and lymph nodes seen in the right cervical region [Figure 2b]. Purified protein derivative (PPD) skin test was positive in two cases and two cases showed a positive family history of tuberculosis. *Mycobacterium tuberculosis* polymerase chain reaction (PCR) was available in one patient and was positive. On fine-needle aspiration cytology (FNAC) examination, all cases showed epithelioid granuloma and caseous necrosis in the background of inflammatory infiltrate [Figure 3a and b]. On the basis of above findings, all patients were diagnosed as tubercular parotitis and were treated with antitubercular multidrug regimen

- Group 2: Recurrent swelling with fistula: This group consisted of one patient, who was a 30-year-old male presented with recurrent swelling and watery discharge from an opening over the left parotid region in the last 3 months. There was no associated pain or fever. The patient briefed the past history of swelling over the left parotid area for which he consulted a general practitioner and got incision and

drainage and at that time, got symptomatic relief for 3–4 months. However, later on, he developed on-and-off watery discharge for which the patient was referred to us. On clinical examination, there was a single 3.5 cm × 3.0 cm swelling in the left parotid region which was firm, mobile, and nontender with chronic discharging sinus over it. Facial nerve function was normal. There was no cervical lymphadenopathy. Routine hematological investigation and chest X-ray were normal. The patient underwent superficial parotidectomy with uneventful postoperative recovery [Figure 4a]. On gross inspection, surgical specimen was covered with skin flap which measured 5.0 cm × 4.0 cm × 2.5 cm [Figure 4b]. Cut surface showed salivary gland parenchyma with small foci of necrosis. Histopathological examination revealed multiple granulomas with central caseation, destroying the gland parenchyma. Periglandular lymphoid follicles also showed similar granulomas and necrosis merging into the surrounding fibrous stroma,



Figure 1: Tender and soft swelling in the left preauricular region (a). Nontender and firm swelling in the left preauricular region (b)

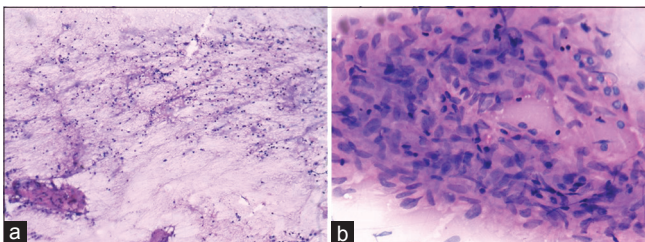


Figure 3: Epithelioid granuloma in the background of caseous necrosis and inflammatory cells (H and E, ×100) (a). Granuloma in high-power view (H and E ×400) (b)



Figure 2: Diffuse and firm swelling in the right preauricular region (a). Computed tomography scan of the same patient showing soft-tissue attenuation in the deep lobe of the right parotid gland (b)

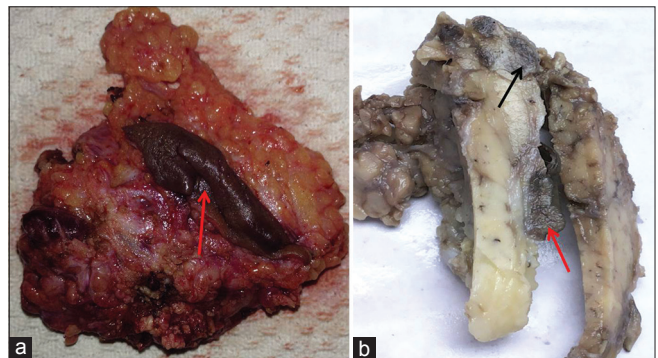


Figure 4: Excised specimen with fistulous tract (arrow) and surrounding skin (a). Gross specimen: Cut surface of parotid gland showing gray-white gland parenchyma with foci of necrosis (arrow) (b)

**Table 1: Summary of clinical and pathological findings of cases**

Details	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8
Age/sex	47/female	65/male	28/male	18/male	30/male	16/female	12/male	27/male
Site of lesion	Right	Left	Left	Left	Left	Left	Right	Right
Duration	4 months	3 months	6 months	2.5 months	8 months (recurrent)	25 days	5 months	<1 month
Description of the mass	3 cm × 3 cm, soft to firm, mobile, nontender	4 cm × 3.5 cm, soft to firm, mobile, nontender	2 cm × 2 cm, soft to firm, mobile, nontender	1.5 cm × 1.5 cm, soft to firm, mobile, nontender	3 cm × 3 cm, firm, mobile, nontender, with small discharging cutaneous fistula	2.5 cm × 2 cm, soft, mobile, and tender	2 cm × 2 cm, soft to firm, mobile, nontender	2 cm × 2.5 cm, firm/tender
Chest X-ray	Chest X-ray normal	Chest X-ray normal	Chest X-ray normal	Chest X-ray normal	Chest X-ray normal	Chest X-ray normal	Not available	Not available
PPD test	Negative	Negative	Positive (28 mm)	Positive (42 mm)	Not done	Not done	Positive (32 mm)	Positive (30 mm)
Other associated findings	CT scan hypodense area suggesting necrosis in deep lobe of parotid gland	<i>Mycobacterium tuberculosis</i> PCR-positive	Upper small cervical lymphadenopathy (<1 cm), history of close contact in family		History of close contact with tuberculosis in younger brother	Upper small cervical lymphadenopathy (<1 cm)	History of close contact in family	No specific finding
Procedure done and material obtained	FNAC - blood-mixed pus	FNAC - blood-mixed dirty fluid	FNAC - dirty cheesy material	FNAC - dirty cheesy material	Histopathology superficial parotidectomy with removal of fistulous tract)	FNAC - pus aspirated	FNAC - Whitish material	FNAC - blood-mixed pus aspirated
Pathological findings	Caseous necrosis and epithelioid granulomas	Epithelioid granulomas and mixed inflammatory cells	Caseous necrosis and epithelioid granulomas	Abundant caseous necrosis and epithelioid granulomas	Abundant caseous necrosis and epithelioid granulomas, granulation tissue	Caseous necrosis and epithelioid granuloma with acute inflammatory cells	Necrosis and epithelioid granuloma	Epithelioid granuloma, acute inflammatory exudate with focal necrosis, few benign acinar cell cluster
Ziehl-Neelsen stain for AFB	AFB Not detected	AFB detected	AFB Not detected	AFB Not detected	AFB detected	AFB detected	AFB Not detected	AFB Not detected

FNAC: Fine-needle aspiration cytology, PCR: Polymerase chain reaction, AFB: Acid-fast bacilli, PPD: Purified protein derivative

with granulation tissue extending up to the skin (formation of fistulous tract). Granuloma was comprised of epithelioid cells, lymphocytes, and Langhans giant cells and rimmed by fibroblasts [Figure 5a-f]. On the basis of the above histomorphological findings, this case was diagnosed as tubercular parotitis. Antitubercular treatment (four-drug regimen) for 9 months was given and the patient has no recurrence or any complaint on 9-month follow-up

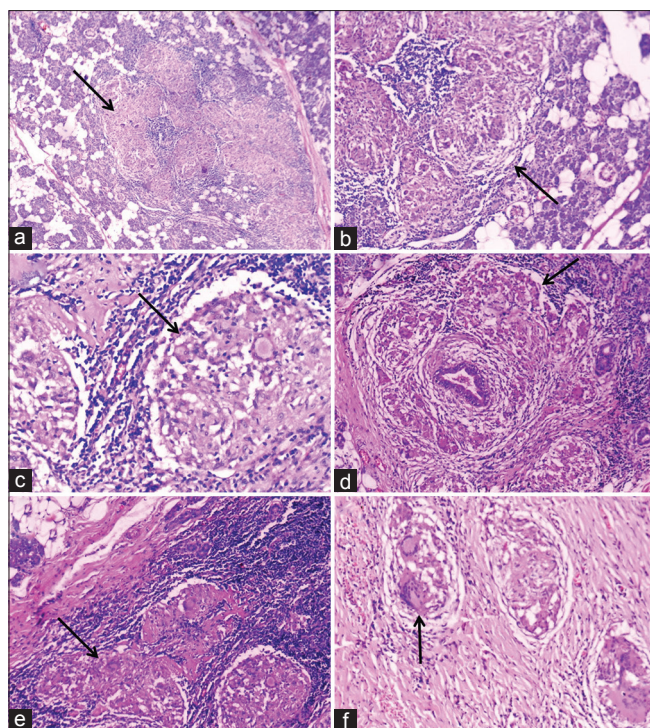
- Group 3: Acute inflammatory swelling/abscess: Consisted of two cases: a 16-year-old female [Figure 1b] and a 27-year-old male presented with painful preauricular swelling for <1 month. There was no history of fever and other systemic symptoms. Clinical examination showed soft, mobile, tender, and indurated swelling. Routine biochemical and hematological measurements and chest X-ray were normal. On FNAC, pus was aspirated. Microscopy showed multiple epithelioid granuloma and caseous necrosis in the background of acute inflammatory exudates and hence cases were diagnosed

as tuberculous abscess. Both patient showed complete resolution of preauricular swelling after anti-tubercular drug therapy.

## DISCUSSION

The incidence of tuberculosis is increasing among the general population of developing countries. In spite of being a common infection, tuberculosis of major salivary glands is rarely seen. Parotid tuberculosis constitutes 2.5%–10% of salivary gland tuberculosis.<sup>[6,7]</sup> Lee and Liu studied the largest case series which consisted of 49 patients of tubercular parotitis and described the clinicopathological feature of this rare entity.<sup>[8]</sup> Salivary glands are believed to resist the growth of tubercular bacilli due to continuous flow of saliva which prevents the lodging and growth of bacilli, while thiocyanate ions and various proteolytic enzymes impart the antibacterial property.<sup>[2,6,8]</sup>

The way *Mycobacterium* reaches the parotid gland is still not very clear. Involvement of the parotid gland and its lymph



**Figure 5: Histopathology: Multiple epithelioid granuloma in parotid gland parenchyma (H and E,  $\times 40$ ) (a). (H and E,  $\times 100$ ) (b). (H and E,  $\times 400$ ) (c). Periductal granuloma (H and E,  $\times 200$ ) (d). Periglandular lymphoid tissue involvement by granuloma (H and E,  $\times 200$ ) (e). Periglandular fibrous tissue showing granuloma with Langhans giant cell (H and E,  $\times 400$ ) (f). (granuloma shown by arrow)**

nodes may develop in two ways: first, a focus of mycobacterial infection in the oral cavity (infected sputum, tonsil) liberates bacilli that ascend into the salivary gland via its duct or pass to its associated lymph nodes via lymphatic drainage. The second pathway involves hematogenous or lymphatic spread from a distant primary lung focus.<sup>[5,7,9]</sup> In the current study, three parotid swellings (3/8 cases) were associated with lymphadenopathy (in one case, it was too small that was only detected on CT neck). Thus, association with cervical lymphadenopathy in these patients further strengthens the theory of lymphatic spread.

Clinical presentation varies from an acute to chronic sialadenitis. Chronic form mimics tumorous condition, where patient presents as slow-growing asymptomatic painless mass with or without cervical lymphadenopathy. Radiology is also nonspecific. Sometimes, diagnosis can be a histological surprise.<sup>[10]</sup> In acute form, there is diffuse involvement of gland parenchyma by disease for short duration mimicking acute inflammatory lesion (abscess). In this study, 2/8 cases presented as acute inflammatory lesion with a short history [Figure 1b]. It may also be presented as a periauricular fistula.<sup>[8-13]</sup>

The most common involvement is unilateral but occasionally bilateral also.<sup>[8]</sup> It is found in both males and females equally. Tubercular parotitis presents as two histomorphological

types. The localized type shows the involvement of pericapsular or intracapsular lymph nodes by caseating or noncaseating granulomas first and then it gradually spreads to the glandular parenchyma. The other type is rare and shows predominant and diffuse involvement of salivary gland parenchyma initially.<sup>[3-8,13]</sup> In the third case of this study, histopathology showed predominant glandular involvement by multiple well-formed caseating granulomas and necrosis, destroying the glandular architecture and also around the ducts [Figure 5a-d]. Pericapsular areas were also involved by granulomas and necrosis which were extending to the subcutaneous plane. This patient had a previous history of incision and drainage which thereafter resulted into formation of the fistulous tract. Also in suspected cases of tubercular abscess, incision and drainage should not be done.

The definitive diagnosis is made by FNAC or histological examination after parotidectomy.<sup>[3,10-13]</sup> Sometimes, tuberculous parotitis becomes a diagnostic challenge in the absence of clinical disease in lung and it requires a high degree of clinical suspicion and adds of other supportive investigations. A chest radiograph may be helpful in cases of associated pulmonary tuberculosis. None of our patients have chest radiographic evidence of either active or prior pulmonary tuberculosis. The use of erythrocyte sedimentation rate and tuberculin skin testing (PPD test) can provide valuable information, but requires an initial suspicion. In this study, four cases showed positive PPD test ( $> 12$ -mm induration). Although the imaging studies are nonspecific, they may provide a clue to the diagnosis. Ultrasonography is sensitive for lesion situated in the superficial lobe of parotid gland, while CT/magnetic resonance imaging is better for deep-lobe involvement.<sup>[5,8,11]</sup> FNAC is now considered as a useful and reliable technique for the diagnosis of tuberculous parotitis.<sup>[1,7,8,10]</sup> Further, sensitivity is enhanced by utilization of fine-needle aspirate in the detection of *Mycobacterium* using Ziehl–Neelsen staining, culture, and evaluation by PCR.<sup>[9,11,13]</sup> Handa *et al.* reported five cases of tubercular parotitis by FNAC, which is also supported by other authors<sup>[3,7]</sup> However, it has only value when positive.

Tumor together with the tuberculosis in the parotid gland is an extremely rare possibility and we can miss either of two. FNAC may cause false-positive results especially in case of large and necrotic parotid gland neoplasms. Watanabe *et al.* in 2001 diagnosed tuberculous parotitis in postoperative histopathological study, where a patient was operated for adenolymphoma.<sup>[4]</sup> Thus, FNAC should be preferred first in the evaluation of a parotid mass. In addition, it should be also considered that, if there is a doubt in relation to diagnosis, exploratory parotidectomy becomes essential to verify

histological diagnosis.<sup>[2,4,13]</sup> Incisional biopsy or drainage should not be used as it may result in the development of cutaneous fistula as happened in our fifth case.

If the diagnosis is made preoperatively, then patient is spared of surgery and can be cured by medications only. Medical treatment consists of combination antitubercular drugs according to suitable regimen 2RHZ/4RH for a period of 6–9 months.<sup>[2,8,10,11]</sup>

## CONCLUSION

Although tuberculous parotitis is a rare entity, it should be always kept in mind as a differential during evaluation of solitary parotid mass lesion. FNAC and contrast CT imaging technique are reliable tools for early diagnosis and treatment, which can spare patients from complications and surgical intervention.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

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