

## Case Series

# Parotid gland tuberculosis-presentation of 4 case variants

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**Received:** 26 January 2021

**Accepted:** 11 February 2021

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### ABSTRACT

Pulmonary tuberculosis is a necrotizing granulomatous disease with diverse presentation. Nevertheless, extra pulmonary involvement although rare even in endemic regions, is associated with diagnostic dilemma for confirmation and treatment modalities. Contextually, parotid gland tuberculosis is a rare surgical presentation because of nonspecific signs, symptoms and allegedly low incidence; it is often misdiagnosed as parotid neoplasm, and therefore remains a diagnostic paradox. However, clinical suspicion alongside pathological evidence and imaging helps in diagnosing indecisive questionable cases. We present 4 cases of parotid tuberculosis each with different clinical presentation along with clinical suspicion and pathological diagnosis. Parotid tuberculosis is a rare clinical entity and completely curable by category-I anti-tubercular therapy. It has no residual deformity if diagnosed timely followed by conservative treatment management.

**Keywords:** Parotid gland, Tuberculosis, Revised National Tuberculosis Control Program, Acid fast bacilli, Purified protein derivative

### INTRODUCTION

Extra-pulmonary head and neck tuberculosis comprise 10% of TB with cervical lymphadenitis as predominating feature.<sup>1</sup> In India, although there is high incidence of pulmonary tuberculosis, but tuberculosis of parotid gland is uncommon even in tubercular endemic areas. Nonspecific signs, symptoms and clinical presentation combined with low incidence commonly leads to misdiagnosis as neoplasm of parotid gland, owing/ due to limited clinical differentiation and suspicion. FNAC associated with high index of sensitivity (80%) and specificity (93%) along with lymph node involvement, complements to accuracy to FNAC diagnosis. From clinical perspective, localized mass with intra-capsular or peri-capsular presentations alongside sialoadenitis results in diffuse gland involvement which later culminates into parotid abscess.<sup>2</sup> Moreover, clinical diagnosis and Zeil Nelson staining are the minimal investigation may

circumvent unnecessary parotidectomy with added risk of facial nerve injury.<sup>2</sup> Accordingly, for any indecisive undiagnosed cases, exploratory parotidectomy may be mandatory or obligatory to arrive at definite diagnosis.<sup>3</sup> Intriguingly, among extra pulmonary tuberculosis accounting for 20% of tubercular cases, solitary parotid gland involvement is very rare specifically sparing other organs.<sup>3</sup> Current opinions as regards to origin of the disease in parotid gland are speculative, but not limited to the extension of the infection along Stenson's duct from the oral-pharynx, vascular mode of spread from primary focus elsewhere in the body, or through ulcerated oral mucosa.

We report herein four cases of parotid gland tuberculosis being cured in our surgical clinics following presentation with spectrum of clinical features. They were photographed after obtaining due signed consent from them. All cases responded to category I of anti-tubercular

treatment given for six months and recovered completely without any residual facial palsy or parotid gland functional impairment, and currently being followed up for 3 years.

## CASE SERIES

### Case 1

A 45 years old female housewife presented with painless multiple swellings in right parotid region since past 6 months. The swellings were insidious in onset, gradually progressive in size with no history of fever, cough, anorexia, weight loss, night sweats, and tuberculosis exposure in recent past. Clinically, multiple non-tenders, firm swellings in parotid region were noticeable. Oral cavity, cervical lymph nodes and rest of the systemic examination were found to be normal. The swellings corresponded to parotid gland with surrounding redness. No other symptoms and signs of tuberculosis were noted elsewhere in the body. PPD test was strongly positive, ESR was raised (45 mm), FNAC showed epithelioid cell granulomas. Since there were no other evidence focus of tuberculosis, diagnosis of primary parotid tuberculosis was suspected. Patient was put on category I treatment of RNTCP (Revised National Tuberculosis Control Program) for six months. Patient responded to ATT (anti tubercular treatment) with complete resolution.



**Figure 1: (A) Parotid tuberculosis with surrounding redness. (B) Positive PPD test.**

### Case 2

A 21 years old male presented with inability to open mouth and with facial asymmetry. There was no history of injury, fever, breathing difficulty, weight loss, tuberculosis or exposure to tuberculosis in the recent past. On examination, there was a fluctuating cold abscess of size 4x5 cm in the right parotid region. Systemic examination was normal with no evidence of tuberculosis elsewhere in the body. X-ray of the tempo-mandibular joints was normal. Antigravity aspiration of pus was sent for culture for pyogenic organism and tuberculosis. The culture yielded no growth after 48 hours of incubation. Repeat antigravity aspiration done twice relieving the patient of trismus. Aspirate sent for AFB staining was found to be positive, also PCR done from the aspirate

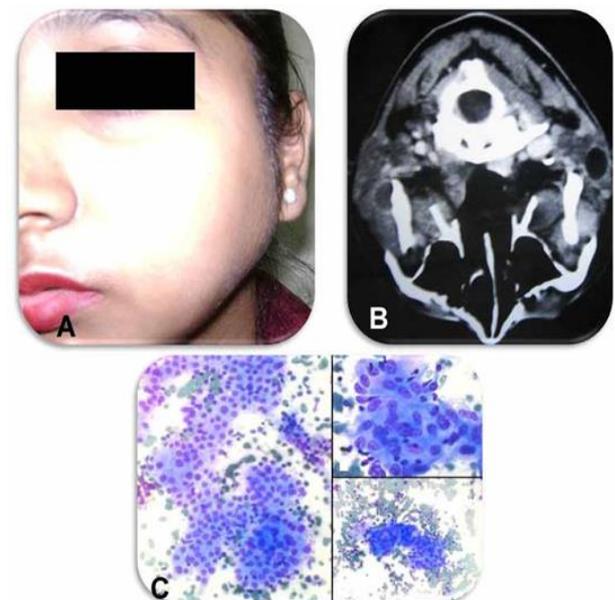
clinched diagnosis of parotid tuberculosis. Diagnosis was supported by strongly positive PPD test. The patient was given a course of anti- tubercular treatment (category I) which led to complete resolution of the symptoms with no restriction of jaw movements.



**Figure 2: (A): Cold abscess R parotid gland. (B): Restricted jaw movements (C): AFB on ZN staining.**

### Case 3

A 28 years old female presented with diffuse swelling of the left parotid region causing facial asymmetry. No other signs and symptoms suggestive of tuberculosis elsewhere in the body were found. On examination, there was diffuse swelling of left parotid gland which was non-tender, firm with normal overlying skin. Oral cavity, oral pharynx, TM joints and ears were normal with no evidence of cervical lymphadenopathy. Systemic examination was unremarkable. The patient was subjected to blood, radiological and pathological investigations. PPD test was found positive. ESR was 54 mm. CECT head and neck showed central necrosis in left parotid with collection. FNAC done for confirmation of diagnosis showed epithelioid cell granulomas and Langhan's type giant cells. Category I ATT given for 6 months led to complete resolution of symptoms.



**Figure 3: (A) Diffuse swelling L parotid gland, (B) Central necrotic area, (C) Epithelioid cell granuloma.**

#### Case 4

An 18-year-old female developed facial asymmetry with mild restricted mouth opening and diffuse swelling of right parotid gland of three weeks duration with no systemic disease, with no family history of tuberculosis or exposure. Inflammatory skin changes with mild tenderness suggestive of parotitis and asymmetry of face with mild restriction of jaw movements was noted suggestive of chronic parotitis with no regional lymphadenitis. ESR was 60 mm first hour and PPD was strongly positive with central necrosis. FNAC was granulomatous disease suggestive of tuberculosis. She responded initially to anti-inflammatory drugs with restoration of jaw movement and completely responded to category I anti-tubercular treatment.



**Figure 4: (A) Diffuse swelling; (B) Limitation of jaw movements; (C) PPD positive with central necrosis.**

#### DISCUSSION

All 4 cases had common presentation of diffuse swelling localized to the region of parotid gland, gradually increasing over time. Additionally, superadded infection and cold abscess may contribute to pain and facial asymmetry. Limitation of jaw movement was encountered in two of our cases. Intriguingly, opposite parotid gland was found normal upon examination in all 4 cases. According to Iseri et al diffuse swelling may contribute to facial asymmetry may progress to abscess or fistula without any constitutional symptoms and the CT imaging obtained with or without contrast may reveal homogenous enhancement.<sup>4</sup>

Benign parotid tumor is a common misdiagnosis for tubercular parotid gland and high index of suspicion is the clue to diagnosis.<sup>5</sup> Radiological investigations provide clue and USG is better when superficial lobe is involved; CT scans has limited non-specific findings, but MRI helps in delineating deep lobe involvement.<sup>5</sup> Routine simple tests such as FNAC followed by Giemsa or ZN staining can distinguish chronic parotid swelling with acute or chronic suppurative infection.<sup>6,7</sup> Moreover, inconclusive FNAC and Imaging studies may allow surgeon to opt for surgical intervention as Watanabe in 2001 operated preoperatively diagnosed case of adenolymphoma which on histopathological examination was confirmed as tuberculosis of parotid gland.<sup>8</sup>

Handa et al reported five cases of tuberculosis of parotid gland diagnosed by FNAC and managed conservatively.<sup>9</sup> Another seven out of eight cases of parotid tuberculosis presenting as diffuse swelling harboring acute parotid inflammation and diagnosed with FNAC, responded completely to anti tubercular therapy; while, one case with unilateral recurrent swelling in pre-auricular region with fistula required superficial parotidectomy with excision of fistula track.<sup>10</sup> Incision and prior drainage of cold abscess results in fistula formation and must be avoided.<sup>10</sup> As suggested above, initial evaluation of any parotid mass, FNAC is highly sensitive (81-100%) and with specificity of (94-100%).<sup>4</sup> Furthermore, in inconclusive cases; FNAC in cases of necrotic neoplasm, culture of aspirate with suspicion of tuberculosis is warranted.<sup>11</sup>

MRI is of diagnostic help when invasion of multiple sites of parotid gland or peri-parotid gland invasion occurs.<sup>5</sup> A diffuse solitary swelling of unilateral parotid gland without any evidence of tubercular lesion elsewhere is difficult to diagnose but clinical suspicion of this rare presentation goes a long way in avoiding surgical interventions, as they finally respond to conservative anti tubercular treatment.<sup>5</sup>

A thirty-five-year-old lady presenting with painful right parotid lump was taken up for superficial parotidectomy. Her histopathological report specifying granulomatous intra-parotid lymph node involvement without any necrosis was suggestive of tubercular involvement of parotid gland.<sup>12</sup> PPD was strongly positive (>20 mm) and she responded to 9 months of anti-tubercular treatment without any residual deformity.<sup>12</sup> As emphasized by others, any differential diagnosis of mixed parotid tumor, lymphoma, chronic lymphadenopathy, sialosis, sjogren syndrome and acute with chronic suppurative parotitis should not be overlooked as AFB may not always be seen in biopsy specimen due to very low concentration of bacteria in extra-pulmonary lesions.<sup>12</sup>

Gradually progressive swelling of right parotid gland of 6 months duration has been reported in an 38 year old lady diagnosed by FNAC revealing lymphocyte, histiocytes, epitheloid cells forming granulomas with multi nucleated giant cells in caseous necrotic background.<sup>13</sup> The tuberculin skin test was 38 mm in 72 hours with vesicle formation.<sup>13</sup> Bilateral parotid gland tuberculosis may reportedly result from direct infected source such as tonsils, infected tooth, retrograde by duct or haematogenous or lymphatic spread from lungs.<sup>14</sup>

Parotid tuberculosis has also been reported from post renal transplant patient on Prograft (FK506) immunosuppressant, developing painful swelling of left parotid-masseter region of 2 months duration with no systemic sign of tuberculosis elsewhere including sputum culture for AFB. USG revealed multiple tender hypoechoic nodular areas suggestive of superficial lymphadenitis. Aspiration cytology revealed caseous pus,

positive for tubercular bacilli with ZN stain and confirmed by RT PCR.<sup>15</sup>

Of noteworthy interest was our findings that simple investigations such as PPD reading of >12 mm in size together with ESR and FNAC as investigation confirmed diagnosis in all 4 investigated /reported cases without need for any surgical interventions.

## CONCLUSION

Parotid tuberculosis is an infrequent disease, but clinical suspicion and investigations can help in diagnosis. Judicious and conservative management with anti-tubercular treatment can defer fistula formation. All 4 patients were followed for last 3 years and no residual deformity, recurrence or involvement of opposite gland have been noticed till date.

*Funding: No funding sources*

*Conflict of interest: None declared*

*Ethical approval: Not required*

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**Cite this article as:** Paruthy S, Paruthy SB. Parotid gland tuberculosis-presentation of 4 case variants. *Int Surg J* 2021;8:984-7.