Case Report

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Unusual Presentation of Extra Pulmonary Tuberculosis in Middle aged Female: A Unique Case Report.

Madeeha Shahid, Pirhay Fatma, Nazia Qamar, Muhammad Salman Zafar, Aun Ali, Aamir Hussain

ABSTRACT:

Pulmonary tuberculosis (TB) is a leading cause of morbidity and mortality among all infectious diseases especially in Pakistan where TB is endemic while extra pulmonary TB often encountered. However, primary salivary gland specifically unilateral parotid gland involvement is exceedingly uncommon. We present a case of 38-year old woman with six months duration of right parotid lump. The lump was firm and non-tender with bilateral cervical lymphadenopathy. No other constitutional symptoms of TB in our patient except weight loss. We presumed it a parotid neoplasm but after workup it reveals parotid TB which responds to first line anti-tuberculous therapy (ATT). So, careful exclusion of parotid neoplasm prevented unwanted surgery in our patient and patient recovered without any residual disease. We proposed that the differential diagnosis of a parotid gland swelling should include the infrequent possibility of parotid gland tuberculosis.

Keywords: Anti-tuberculous therapy, Extra-pulmonary Tuberculosis, Fine needle aspiration cytology, Parotid tuberculosis.

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BACKGROUND

TB is a chronic necrotizing granulomatous disease caused by Mycobacterium tuberculosis primarily affecting the lungs known as Pulmonary Koch's. TB is endemic in Pakistan with the incidence of 264 per 100,000 people. Out of which 20% of cases have extra-pulmonary presentation[1]. TB rarely involves salivary glands with less than 200 cases of parotid TB reported yet[2]. The involvement of the salivary glands is even more uncommon because the constant flow of saliva prevents tubercular bacilli from building up in them. Owing to slow salivary flow, the parotid glands are more susceptible to be affected than other salivary glands[3]. Diagnostic challenges arise from the fact that parotid gland TB presents similarly to a parotid tumor[4], and also when there is no relevant clinical history of TB.

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CASE PRESENTATION

A 38-year-old housewife presented at the outpatient clinic with the complaint of a rounded painless swelling below and in front of the right ear lobule for last 6 months, swelling was insidious in onset and gradually increased in size and attained a size of 3 x 3 cm. It was not associated with pain or change in size while chewing food. No history of asymmetry of face, difficulty in closing eyes, difficulty of chewing food, or drooling of saliva from mouth. No history of other swelling. There was a history of weight loss. She lost 6 kg over these months. She had no prior surgery or hospital admission. She was known hypertensive for which she has been taking verapamil for last 2 years. No history of autoimmune disease, mumps in childhood, earache, fever, and dental infection. No history of other symptom like fever, malaise, anorexia, cough, and discharge from swelling except her family history was positive for pulmonary tuberculosis, and there was no history of addiction or smoking.

On examination, there was a well-defined smooth surface spherical swelling of about 6 x 5 cm on right parotid region displacing right ear lobule upwards and outwards, it was firm in consistency with no signs of inflammation. The

swelling was not fixed to the underlying masseter or overlying skin. No sign suggestive of facial nerve palsy. Oral examination was unremarkable with normal parotid duct opening. There were multiple mobile right sided cervical lymph nodes.

On routine blood test, her total leukocyte count was 18.4 (Normal Range 4-11 x 10^3 /ul) with predominately lymphocytosis, Erythrocyte Sedimentation Rate was 80 mm/hour.

Her ultrasound neck showed an enlarged right parotid gland having a heterogenous texture with small hypoechoic focus, on Doppler increased vascularity was seen and this suggested parotitis. And there were multiple subcentimetric right cervical lymph nodes.

Her contrast enhanced CT scan (CECT) from base of skull to root of neck showed a mildly enlarged parotid gland without any necrosis and calcification, and no evidence of sialolithiasis. Multiple subcentimeter right cervical lymph nodes were seen. (Figure: 1). Fine needle aspiration cytology (FNAC) of right parotid gland and cervical lymph node both were carried out. FNAC of right parotid showed acellular smears comprising necrotic debris showing a caseous appearance. (Figure: 2). FNAC of right cervical lymph node (LN) showed cellular smears comprising two small collections of epithelioid histiocytes, having lymphoid cells in varying stages of maturation. (Figure: 3). It raised suspicion of tuberculosis so an excisional biopsy and high-resolution computed tomography (HRCT) of the chest was performed to rule out pulmonary tuberculosis. Excisional biopsy of cervical LN showed chronic granulomatous inflammation with necrosis. (Figure: 4). Her HRCT of the chest showed few calcified sub-centimeter-sized mediastinal and bilateral hilar lymph nodes.

Excluding benign and malignant growth and other infective etiologies patient was diagnosed with Primary Tuberculosis of unilateral Parotid gland involving cervical LN without lung involvement.

Meanwhile, Mantoux test became positive (30 mm in 72

Figure: 1. CECT scan shows only Right-sided Parotid gland enlargement and contralateral side is normal

Figure: 2. FNAC of right Parotid shows Caseous necrosis.

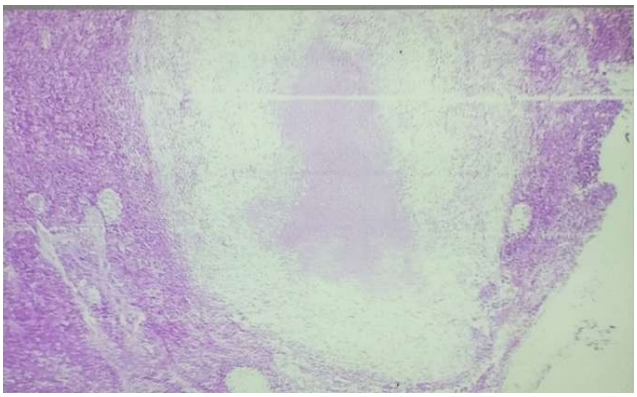


Figure 3: FNAC of Cervical Lymph node shows epithelial histocytes.

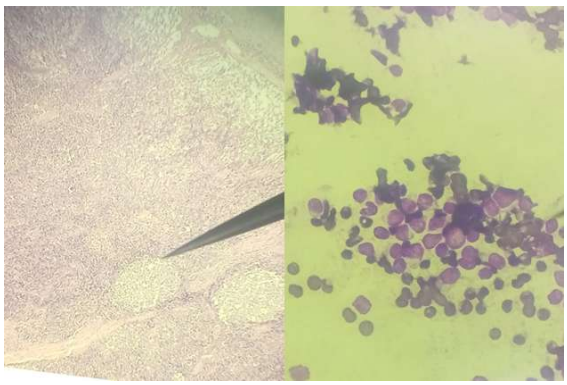
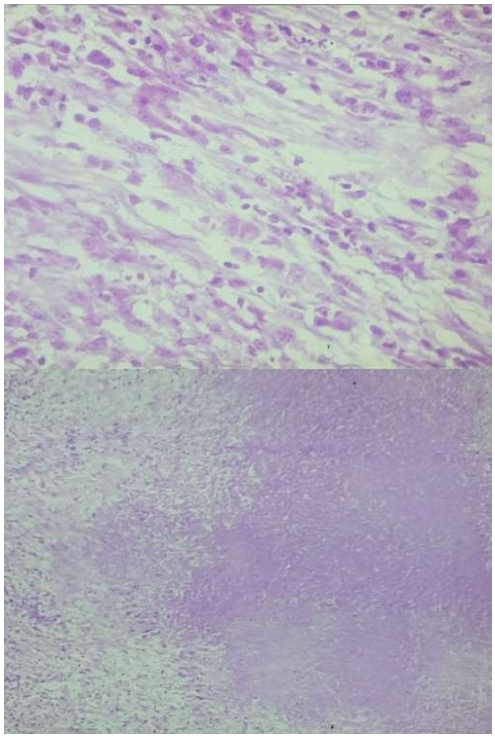


Figure: 4. Excision Biopsy of Lymph node shows Chronic Granulomatous inflammation.



hours with vesicle formation) which strongly support our diagnosis. The patient was prescribed first line ATT for 1 year with liver function monitoring and counseling for strict compliance of therapy. Result was satisfactory.

DISCUSSION:

The long-known cause of chronic necrotizing granulomatous disease is Mycobacterium tuberculosis. Ten percent of extra-pulmonary sites are in head and neck area, with cervical lymph nodes being the most frequently affected. The larynx, deep neck space, and otitis media are the next most frequently affected regions. The skin, mouth cavity, oropharynx, nose, thyroid, salivary glands, and mandible are some less frequent sites. The first case of secondary parotid TB was documented in 1893. And the first case of primary parotid TB was reported in 1894, a year later [3]. Even in Pakistan, where tuberculosis is ubiquitous, salivary gland involvement is uncommon. The constant flow of saliva and presence of proteolytic enzymes like lysozymes and thiocyanate ions in salivary gland secretions may be the cause of this[4].

Parotid TB is comparatively more common in males[5], but in our case it found in female. Rarely does the localization occur in both parotid glands simultaneously; it is typically unilateral[8]. The majority of cases of parotid TB occur in adults, with a median presentation age of 45 years[5]. Parotid tuberculosis does not have any particular symptoms or clinical signs; it typically manifests as a slowly growing painless parotid lump[5,8] mimicking benign parotid tumor[6]. Sometimes, fistulization of the swelling can lead to the direction of tubercular pathology, but most of the time cutaneous plane remains intact with minimal or no evidence of inflammation. Rarely, tubercular sialadenitis results in trismus. Since parotid swelling is linked to cervical adenopathy, it may happen on its own. It is rare for primary disease to exhibit tubercular symptoms such as fever, asthenia, weight loss, and nocturnal sweats[8]. Parotid abscess is another possible presentation. Long-standing parotid abscess incision and drainage should delay until tuberculosis has been ruled out since doing so could cause a fistula or sinus formation[7]. In 2022, the first incidence of bilateral parotitis with facial nerve palsy was documented by Jameel Z et al which are extremely rare presentation of primary parotid TB[9].

Along with FNAC, ultrasonography and CECT are effective methods for diagnosis confirmation; however, the FNAC report may not be definitive if the infection is active. Better findings are obtained when aspirated material is stained with AFB in conjunction with cytological studies[7]. In cases where FNAC and imaging yield conflicting results, surgical intervention is required to collect tissue for histological analysis. Typically, an excisional biopsy is carried out; however, if the entire parotid is affected, a total parotidectomy can be necessary[7]. There are two pathological typesof parotid tuberculosis based on histology: the diffuse form and the nodular or circumscribed form. The nodular type involves either periglandular or intraglandular lymph nodes. A cyst or cold abscess could be the appearance of it. This variety is more frequent. Pathological alterations may occur in glandular or interstitial tissue. The less frequent diffuse variety is characterized by both large and small caseation or abscesses that affect the gland parenchyma as a whole[5,7].

The only course of treatment after diagnosis is ATT. Belatik H et al. in 2018 report two cases of parotid TB on histology after excision of parotid gland[8]. But in our case, we diagnosed on FNAC and antimicrobial treatment alone is beneficial, allowing the parotid swelling to subside and the tubercular focus to be eliminated. So, use of parotidectomy for diagnostic and/or therapeutic purposes is no longer a hot topic. For drug-sensitive tuberculosis, the World Health Organization suggests a six-month therapy regimen. The recommendations also apply to extra-pulmonary tuberculosis, with the exception of TB of the central nervous system, joint, or bone[3].

CONCLUSION:

Because of its rarity, diagnosis of parotid TB needs high level of suspicion[1]. It diagnosed accurately with the help of microbiology, histopathology and radiology[1,5,9]. The diagnostic accuracy of FNAC is very high with sensitivity of 81-100% and specificity of 94-100%[6,7,10]. After definitive diagnosis parotid TB is completely curable with ATT[5,6,9]. As in our case, patient is recovered and enjoying happy healthy life. So, no need of unwanted parotidectomy to increase morbidity of patient.

Authors Contributions:
Madeeha Shahid: Research conception, data collection and Writing of final draft
Pirhay Fatma: Research Concept, Data collection
Nazia Qamar: Data Collection
Muhammad Salman Zafar: Data Collection
Aun Ali: Review of Final draft
Aamir Hussain: Review of Final Draft

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