



Bilateral Tuberculous Parotitis with Facial Nerve Palsy: The First Case Report

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Abstract

Tuberculosis of salivary glands is a rare condition, even in endemic countries like Pakistan. Tuberculosis of the parotid gland is even rarer and with cases of bilateral tuberculous parotitis being even more unique. For the first time in literature, we report a novel case of bilateral tuberculous parotitis with facial palsy in a thirty-eight-year-old female with no previously known comorbidities. The female presented with bilateral painful parotid swelling and a long history of constitutional symptoms. Diagnosis of tuberculous parotitis was confirmed with ultrasound-guided fine needle aspiration cytology which revealed variable sized granulomas containing epithelioid cells, macrophages and mature lymphocytes and a positive acid-fast bacilli smear. Patient was treated with anti-tubercular drugs (Isoniazid, Rifampicin, Pyrazinamide, Ethambutol) to which she responded well and after three months of therapy her swelling and facial weakness improved significantly. In addition to reporting a completely new kind of presentation, this paper reviews relevant literature and stresses the need for a high degree of suspicion for tuberculous parotitis in a patient with chronic parotid swelling. Use of fine needle aspiration is important so that this curable condition can be diagnosed and medically treated. In this way, surgical intervention can be avoided and further complications can be prevented.

Keywords: Tuberculous parotitis; Tuberculosis; Parotid gland; Facial nerve palsy

Introduction

Tuberculosis is a contagious disease that mainly infects the lungs but can have extrapulmonary manifestations as well. Among all the cases of tuberculosis, only 25% are extrapulmonary infections, of which only 10% are in the head and neck region [1,2]. It is an established fact that tuberculosis is a major health concern affecting around ten-million people a year and more so, in developing countries like Pakistan, where it is exceedingly prevalent. But even in endemic countries, tuberculous parotitis is rarely reported. A mere two hundred cases of tuberculous parotitis have ever been reported in literature and most of these are of unilateral tuberculous parotitis, diagnosed from post-parotidectomy specimens [3,4]. Bilateral tuberculous parotitis is an even rarer entity and has been reported only six times in literature [1]. Here we discuss a case of bilateral tuberculous parotitis associated with facial palsy. To the best of our knowledge, this is the first such case that is being reported. In addition, the relevant literature about tuberculous parotitis is reviewed.

Case Presentation

A thirty-eight-year-old non-smoker woman with no previously known comorbidities presented with bilateral swelling of the preauricular area for two months. Low-grade fever, cough and weight loss of 8 kg in the past two months was also reported. There was no substantial change in the size of both swellings. Although swellings were initially non-tender but later became painful. There was no associated dysphagia or hoarseness of voice.

On local examination of the parotid region, two firm, non-mobile, tender swellings were present. Left sided swelling was just beneath the mandibular angle measuring 2 cm × 2 cm and right sided swelling was antero-inferior to the right ear lobe measuring 2 cm × 1 cm. No discharging sinus was present (Figure 1).

Associated findings included no wrinkling on the left side of forehead, incomplete closure of the left eye, loss of left nasolabial fold and slanting angle of mouth towards the left side for the past two weeks. All these findings were consistent with left sided facial palsy Grade 3 (House-Brackmann Grading) (Figure 2) [5].

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Figure 1: Bilateral parotid swelling.

General physical and systemic evaluation was unremarkable including examination of the respiratory system. No abnormality was exhibited in routine hematological and biochemical profile including urine routine examination, electrocardiography and ultrasound abdomen. ANA was negative and serum ACE levels were within normal values thus excluding autoimmunity and sarcoidosis as causes of chronic parotitis. Viral serology including HIV was also negative.

Ultrasound of parotid region showed bilaterally enlarged parotid glands (left > right) and heterogeneous echotexture with hypochoic nodules located in the peripheral zone of the hyperechoic parotid gland with markedly enhanced vascular supply on Doppler ultrasound. Rests of the salivary glands were unremarkable. Bilateral multiple small volume cervical and submandibular lymphadenopathy was also noted (Figure 3).

CT-Scan of the neck illustrated left parotid swelling measuring 3.9 cm × 1.8 cm just inferiorly to the angle of mandible and right parotid swelling measuring 3.2 cm × 2.0 cm superficial to the body of mandible in the preauricular region (Figure 4). CT-Scan of the chest revealed bilateral hilar, mediastinal and supraclavicular lymphadenopathy with no active pulmonary infiltrates but few calcified granulomas were seen in both lungs (Figure 5).

Ultrasound guided Fine Needle Aspiration Cytology (FNAC) of the swellings revealed chronic granulomatous inflammation i.e. variable sized granulomas consisting of epithelioid cells, macrophages and mature lymphocytes suggesting tubercular parotitis. Positive acid-fast bacilli smear of the FNAC sample confirmed the diagnosis of tuberculous parotitis.

Initially four anti-tubercular drugs (Isoniazid, Rifampicin, Pyrazinamide and Ethambutol) were prescribed for two months and then Isoniazid and Rifampicin were continued for the next seven months. The patient's swellings significantly reduced in size after 3 months of therapy along with marked improvement in facial weakness.



Figure 2: Signs of Bells palsy.

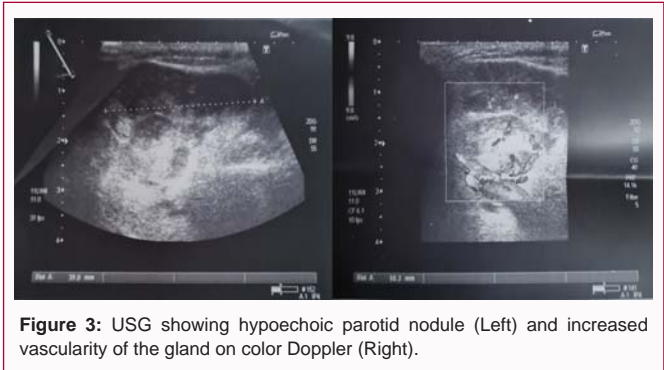


Figure 3: USG showing hypochoic parotid nodule (Left) and increased vascularity of the gland on color Doppler (Right).

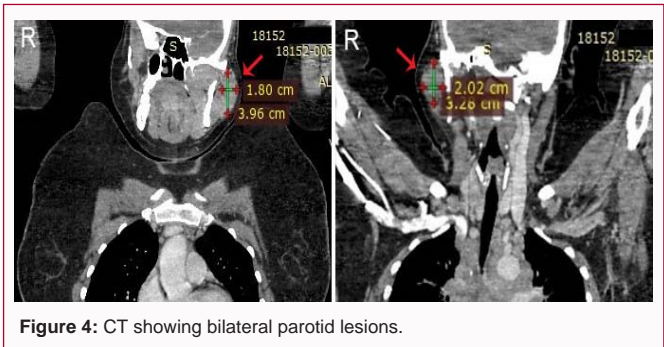


Figure 4: CT showing bilateral parotid lesions.

Discussion

As we have already narrated, tuberculosis most often affects the lungs but can occur anywhere in the body. Among infectious diseases, tuberculosis is a substantial cause of death, second only to COVID-19. It rarely affects major salivary glands and bilateral parotid involvement is even more scarce [6]. In 1983, diagnosis of tuberculous parotitis was first made by C De Paoli [1].

Tuberculous parotitis usually presents as a progressive chronic swelling which can occur *via* ascension of mycobacterium infection from the oral cavity into the duct of the gland or into lymph nodes *via* lymphatic drainage or from a distant primary pulmonary focus *via* hematogenous or lymphogenous route [7]. Tuberculous parotitis is more commonly seen as a secondary infection with its primary source being in the lungs [8]. A total of 25% of patients diagnosed with tuberculous parotitis have an ongoing pulmonary infection [9]. Our patient had tuberculous parotitis with resolved pulmonary infection. Two types of tuberculosis occur in the parotid gland. More common type is intraglandular lymph node involvement and the second less common type being one with parenchymal involvement.

Clinical differentiation of tuberculous parotitis from other

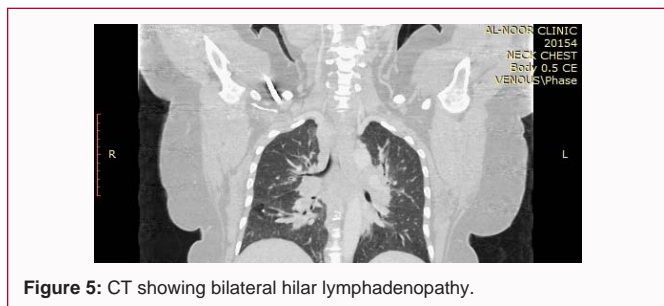


Figure 5: CT showing bilateral hilar lymphadenopathy.

inflammatory diseases poses a significant challenge [7]. Differentials of bilateral parotid swelling are mumps, Sjogren's syndrome, sarcoidosis and malignancy of the parotid gland. Mumps is an acute condition and was excluded in our case by the chronicity of the patient's swellings. Negative ANA and low serum ACE levels helped us to exclude sarcoidosis and other autoimmune diseases especially Sjogren's syndrome. High resolution ultrasonography was used to rule out malignant parotid lesions and played an essential role in the diagnosis of tuberculous parotitis due to its superficial location. It was used to demonstrate whether tuberculous parotitis is of the parenchymal or of the peri-parotid type. Our patient's ultrasound findings included a hyperechoic parotid gland peripherally surrounded by hypoechoic nodules which is consistent with peri-parotid type lesion. It is important to note that radiological investigations are sensitive but not specific in identifying tubercular lesions. Tuberculosis of parotid lesions can be identified by the help of fine needle aspiration cytology with 81% sensitivity and 94% to 100% specificity [2]. Ultrasound guided FNAC of tuberculous parotitis swellings revealed caseating granulomatous epithelioid cell clusters and macrophages characteristic of tuberculosis. Salivary gland acini and malignant cells were not seen in cytology thus excluding malignancy.

Conservative management of tuberculous parotitis is preferred over surgical intervention which is now only used for residual enlarged parotid [10]. Treatment should be started as soon as possible because untreated infections can lead to xerostomia, dental infections and caries, parotid abscess, fistula formation & rarely facial paralysis, septic thrombophlebitis and Frey syndrome. Standard four drug regimen (Isoniazid, Rifampicin, Pyrazinamide, Ethambutol) in the

intensive phase for the initial two months followed by a two-drug regimen (Rifampicin and Isoniazid) in the continuation phase is recommended for treatment of tuberculous parotitis [11].

Conclusion

Tuberculous parotitis is a very rare entity that occurs as a complication of tuberculosis and presents with swelling of the parotid gland. Diagnosis requires a reasonable amount of suspicion with use of radiological and histopathological investigations. Treatment is done with standard antitubercular therapy and prompt start of treatment is necessary to avoid complications.

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