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Parotid gland tuberculosis accompanied by brucellosis

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ABSTRACT

Almost one-fourth of the world's population is latently infected with Mycobacterium tuberculosis (MTB) with approximately 3% to 15% people at risk of progression to active disease during their lifetime. Parotid gland tuberculosis (TB) is an extremely rare form of extrapulmonary TB even in endemic areas. Parotid gland TB presents clinically as a unilateral, slow-growing, and possibly painless mass. Parotid gland TB tends to mimic parotid tumors without pathological evaluation. Risk factors for active infection in extrapulmonary TB forms are human immunodeficiency virus, malnutrition, diabetes mellitus, smoking, alcoholism, hematological malignancies, and immunosuppressive treatments. Brucellosis is a systemic disease that is transmitted from unpasteurized milk and dairy products obtained from an infected animal. It can affect many organs. Brucellosis is difficult to diagnose because its signs and symptoms are nonspecific and mimic many diseases. The aim of this case report is to present the clinical features, pathophysiology, diagnostic process, and treatment of a parotid gland TB case accompanied by brucellosis, the diagnosis and treatment of which were based on the suspicion of the clinician, in the light of the literature.

Keywords: Parotid, parotid gland tuberculosis, brucellosis, QFT-plus, parotid swelling

Introduction

Parotid gland tuberculosis (TB) is an extremely rare form of extrapulmonary TB even in endemic areas (1). Tuberculosis presents clinically as a unilateral, slow growing, and possibly painless mass. Definitive diagnosis can be made only after surgery because it is difficult to differentiate parotid gland TB from parotid gland tumors with clinical and radiological findings (2).

Brucellosis is a zoonotic disease transmitted from animals to humans via unpasteurized milk and dairy products. The disease can be encountered in every region dealing with animal husbandry; however, its occurrence is higher in Mediterranean countries. The disease progresses with nonspecific signs and symptoms (3).

In this case presentation, approach to a parotid mass originating from parotid TB developing on the basis of brucellosis is reported. To the best of our knowledge, this is the first case report presenting such clinical coexistence in the literature.

Case Presentation

A 24-year-old male patient was presented to our clinic with a painless swelling on the right parotid persisting for 2 weeks. He had no known comorbidity. The opposite side was unremarkable. He had not had similar complaints before and had no symptoms of mouth or eye dryness. There was no history of fever, cough, weight loss, or loss of appetite. He also had no history of TB infection or contact with a patient with TB. He was working as a livestock breeder and living in a rural area.

Physical examination revealed a fluctuating swelling of 5×5 cm extending from the parotid tail to the lower jaw. There was no temperature increase or color change on the mass. On performing enhanced computed tomography (CT) scan, we found multiple lesions in the right parotid gland both in the superficial lobe with thin-walled ring enhancement and anterior to the masseter muscle in the deep lobe, with a larger axial diameter of 40×22 mm and fuzzy adjacent fat tissue (Figure 1). In addition, multiple lymphadenopathies (the biggest was 12×8 mm) having hilar echoes were observed near these lesions. Complete blood count results were within normal limits. The erythrocyte sedimentation rate was 39 mm/h, and C-reactive protein was 29.5 mg/dL. HIV test result was negative. Chest x-ray results were within normal limits. The other systems were also found to be normal on examination. Ciprofloxacin 2×500 mg per oral (PO) and metronidazole 4×500 mg (PO) were started

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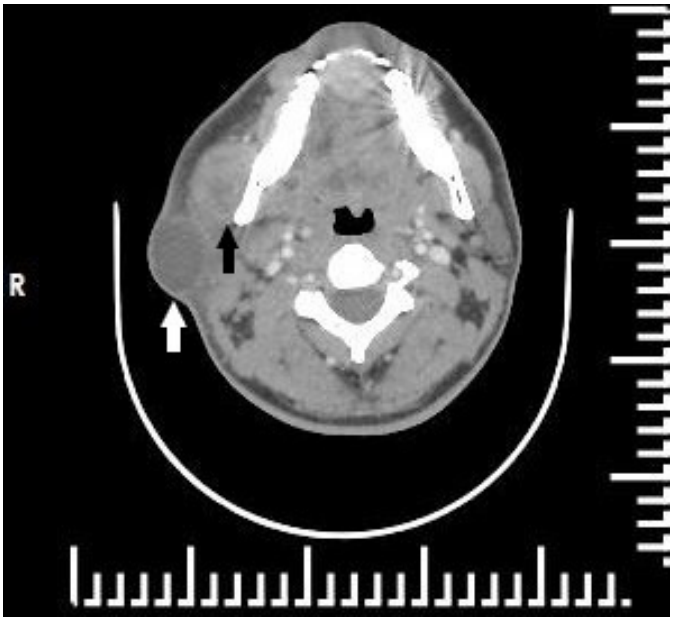

Figure 1. Enhanced computed tomography image shows right parotid gland lesions both in the superficial lobe with thin-walled ring enhancement (white arrow) and anterior to the masseter muscle in the deep lobe (black arrow) with a larger axial diameter of 40×22 mm and fuzzy adjacent fat tissue

empirically considering the infective causes first. On analysis of the abscess drainage fluid, gram stain was negative, no acid-resistant bacilli were found, and no growth was noted on bacterial culture. Histological examination was nondiagnostic. The Rose Bengal test (RBT) was found negative, and standard tube agglutination test (SAT) was found positive at 1/320 titer in peripheral blood samples. On the basis of these findings, antibiotic therapy was changed to doxycycline 2×100 mg PO and rifampicin 2×300 mg PO for systemic brucellosis 1 week after broad spectrum antibiotic therapy was initiated.

Total parotidectomy was performed using intraoperative facial nerve monitoring, because there was no regression in swelling despite the 3-week antibiotic therapy. Although the mass in the superficial lobe was infiltrating subcutaneous adipose tissue, another mass in the deep lobe was attached to the buccal and temporal branches of the facial nerve on the masseter

Main Points:

- Tuberculosis (TB) and brucellosis are diseases that can coexist in people living in endemic areas.
- Brucellosis may be a risk factor for the activation of latent tuberculosis infection and distant organ involvement, such as parotid TB.
- In cases with unilateral parotid swelling, parotid TB should be considered in the differential diagnosis in the presence of risk factors leading to immune system failure.
- The definitive diagnosis of parotid TB is established through the growth of Mycobacterium tuberculosis complex in TB culture and acid-fast bacillus staining.
- In cases where TB bacillus cannot be grown or stained, histopathological examination of necrotizing granulomatous inflammation and Quantiferon-TB Gold Plus test are helpful in diagnosis.

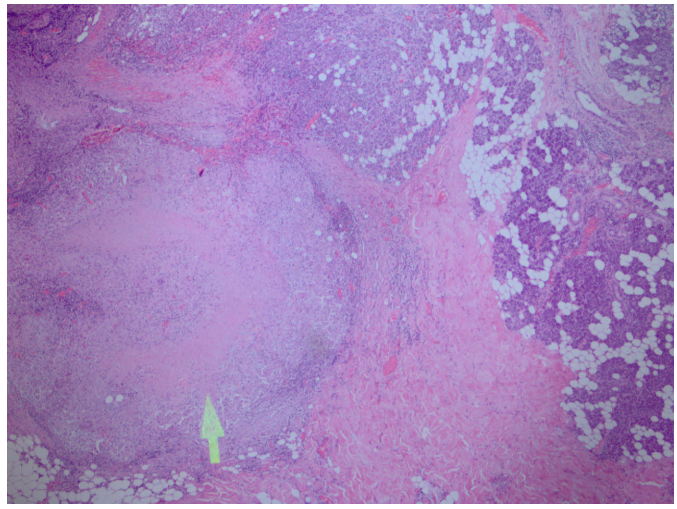

Figure 2. Granuloma structure with large caseous necrosis (arrow) in the middle as well as salivary gland acini under hematoxylin and eosin stain ×100

muscle. The masses were excised along with the deep lobe over the masseter muscle with preservation of all branches of the facial nerve. Granulomatous inflammation with necrosis epithelioid macrophages, Langerhans giant cells, and lymphocytes surrounding the caseous centers were observed on histopathological examination (Figure 2). Histopathological findings indicated TB with a positive Quantiferon-TB Gold Plus (QFT-Plus) test in peripheral blood sample. In the samples collected during surgery, Mycobacterium tuberculosis (MTB) complex grew in the TB (Löwenstein) culture 3 weeks later, whereas Brucella culture was negative. Furthermore, *blood samples* were found to be *positive for Brucella* using SAT (1:640) and RBT, indicating acute brucellosis. On the basis of these results, we interpreted the mechanism of the parotid TB as activation of latent tuberculosis infection (LTBI) in the presence of systemic brucellosis, and a tuberculosis treatment protocol consisting of isoniazid 300 mg 1×1 PO, rifampicin 300 mg 1×2 PO, ethambutol 500 mg 1×3 PO, and pyrazinamide 500 mg 1×4 PO was initiated in addition to doxycycline 100 mg 1×2 PO for brucellosis. One week after surgery, drainage from the parotid area was stopped. There was no evidence of recurrence after a further 6-month follow-up. A written consent form was obtained from the patient.

Discussion

TB is caused by several species of the Mycobacterium tuberculosis complex, including *Mycobacterium bovis*, *M. africanum*, *M. caprae*, *M. orygis*, *M. intracellulare*, *M. canetti*, *M. microti*, and primarily *M. tuberculosis*. Some species of Mycobacterium family are strict pathogens, whereas others are nonpathogenic or opportunistic pathogens. MTB is the major cause of tuberculous cervical lymphadenitis in adults, whereas children are mostly infected by the atypical mycobacteria sp, including *M. avium-intracellulare*, *M. kansasii*, and *M. scrofulaceum* (4).

TB incidence has increased particularly in developing countries owing to increased HIV infection rates, multi-drug resistance against bacteria, and low socioeconomic level in recent years. Although most TB cases are pulmonary, extrapulmonary TB forms account for approximately 25% of all cases. The TB ba-

cilli can present as LTBI without causing disease in individuals with healthy immune systems. Risk factors for active infection in extrapulmonary TB forms are HIV, malnutrition, diabetes mellitus, smoking, alcoholism, hematological malignancies, and immunosuppressive treatments (5). The involvement of parotid gland and intraparotid lymph nodes in TB cases is due to the bacillus being reactivated as a result of decreased immunity in individuals with LTBI and causing distant organ involvement through hematogenic or lymphatic systemic circulation (6). QFT-Plus test, which is a newly introduced in vitro diagnostic laboratory test developed for the detection of TB infections in the whole blood sample, may be useful in differentiating new and distant LTBI and may help physicians make a decision to initiate LTBI treatment (7). Typical radiological findings are not observed in the chest imaging of most patients with parotid TB (8). Parotid gland TB is often characterized with the involvement of intraparotid lymph nodes, and abscess formation is frequently observed in this form of TB (9).

Unilateral parotid swelling generally develops because of bacterial infection, chronic sialoadenitis, duct obstruction, and neoplastic diseases (such as lymphoma and Warthin's tumor). In the differential diagnosis, parotid TB is not considered in the first place when compared with diseases with a similar clinical manifestation. Therefore, majority of the parotid TB cases are operated with the suspicion of malignancy (9). Parotid TB is seen in the form of multiple nodules in CT scan, which is not specific for TB. Necrotizing granulomatous inflammation is observed in histopathological examination. However, granulomatous lymphadenitis does not always indicate TB. The differential diagnosis includes sarcoidosis, berylliosis, toxoplasmosis, syphilis, cat scratch disease, histoplasmosis, coccidioidomycosis, brucellosis, and infection with nontuberculous mycobacteria (10). Nontuberculous mycobacterial infection of the parotid gland is indistinguishable from TB on imaging; however, there is often violaceous discoloration of the overlying skin and purified protein derivative (PPD) skin test result is usually negative or weakly positive in patients with nontuberculous mycobacterial infection (11). It is difficult to show acid-fast bacillus on tissue sections. No growth is observed in MTB culture in general. There is no specific treatment plan defined for the treatment of parotid TB. Complete recovery is achieved with antibiotic therapy protocols used in other organ involvements (12).

Brucellosis is a zoonotic disease caused by gram-negative coccobacilli Brucella spp. It presents with nonspecific findings such as fever, night sweats, fatigue, loss of appetite, and joint pain (13). In most of the patients, it manifests with fluctuating fever and less frequently with the signs of local infection. Axillary or cervical mildly painful lymphadenopathy can be observed in approximately 20% of the cases. Serological tests that can be used in the diagnosis of brucellosis include SAT, RBT, enzyme-linked immunosorbent assay, and immunofluorescence test.

Although the definitive diagnosis is the isolation of the bacteria, a negative *culture does not exclude* the *diagnosis* of *brucellosis* (14). Although there are studies reporting abscess formation in different organs, parotid abscess originating from Brucella was reported only in one case presentation (15).

In conclusion, it should be noted that TB and brucellosis can also involve the parotid gland as well as many different organs, particularly in patients living in endemic areas. In immunocompetent individuals, TB remains dormant without causing disease and may manifest with reactivation in conditions in which immunity is weakened. In this rare case, we saw that acute brucellosis may be a risk factor for parotid tuberculosis with reactivation of the dormant TB bacillus. Therefore, surgery will be useful for both diagnosis and treatment of parotid masses that do not recover despite broad spectrum antibiotic therapy.

Informed Consent: Written informed consent was obtained from the patients who agreed to take part in the study.

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References

1. Lee IK, Liu JW. Tuberculous parotitis: case report and literature review. Ann Otol Rhinol Laryngol 2005; 114: 547-51. [CrossRef]
2. Handa U, Kumar S, Punia RS, Mohan H, Abrol R, Saini V. Tuberculous parotitis: a series of five cases diagnosed on fine needle aspiration cytology. J Laryngol Otol. 2001; 115: 235-7. [CrossRef]
3. Dean AS, Crump L, Greter H, Hattendorf J, Schelling E, Zinsstag J. Clinical manifestations of human brucellosis: a systematic review and meta-analysis. PLoS Negl Trop Dis 2012; 6: e1929. [CrossRef]
4. Hanson RA, Thoongsuwan N. Scrofula. Curr Probl Diagn Radiol 2002; 31: 227-9. [CrossRef]
5. Morán-Mendoza O, Marion SA, Elwood K, Patrick D, FitzGerald JM. Risk factors for developing tuberculosis: a 12-year follow-up of contacts of tuberculosis cases. Int J Tuberc Lung Dis 2010; 14: 1112-29.
6. Hamdan AL, Hadi U, Shabb N. Tuberculous parotitis: a forgotten entity. Otolaryngol Head Neck Surg 2002; 126: 581-2. [CrossRef]
7. Sotgiu G, Saderi L, Petruccioli E, et al. QuantiFERON TB Gold Plus for the diagnosis of tuberculosis: a systematic review and meta-analysis. J Infect 2019; 79: 444-53. [CrossRef]
8. Baldwin AJ, Foster ME. Tuberculous parotitis. Br J Oral Maxillofac Surg 2002; 40: 444-5. [CrossRef]
9. Dixit R, Shah V, Dixit K, Solanki RN, Kotadia J. Tuberculous abscess of parotid gland. J Indian Acad Clin Med 2005; 6:1613.
10. Asano S. Granulomatous lymphadenitis. JClin Exp Hematop 2012; 52: 1-16. [CrossRef]
11. Wei Y, Xiao J, Pui MH, Gong Q. Tuberculosis of the parotid gland: computed tomographic findings. Acta Radiol 2008; 49: 458-61. [CrossRef]
12. Cataño JC, Robledo J. Tuberculous Lymphadenitis and Parotitis. Microbiol Spectr. 2016; 4. [CrossRef]
13. Kaya O, Akcam FZ, Uyar C, Tuz M, Kapucuoglu N. Neck abscess caused by Brucella sp. Infection 2007; 35: 479-80. [CrossRef]
14. Dean AS, Crump L, Greter H, Hattendorf J, Schelling E, Zinsstag J. Clinical manifestations of human brucellosis: a systematic review and meta-analysis. PLoS Negl Trop Dis 2012; 6: 1929. [CrossRef]
15. Yolbaş S, Bozgeyik Z, Artaş G, Koca SS. Parotid abscess secondary to brucellosis in a patient with primary Sjögren's syndrome. Eur J Rheumatol 2018; 5: 62-4. [CrossRef]