

Parotid abscess secondary to brucellosis in a patient with primary Sjögren's syndrome

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Abstract

Swelling in the salivary glands, particularly parotid glands, is observed in approximately one third of Sjögren's syndrome patients. However, such patients should be assessed in terms of causes such as malignancies, infections, amyloidosis, sarcoidosis, and other autoimmune diseases when parotid gland swelling is noted. It should be considered that the incidence of lymphoma increases in Sjögren's syndrome. Unilateral parotid swelling, especially if accompanied by severe pain and redness, should be monitored for suppurative bacterial infections. Brucellosis is a systemic disease that may involve one or multiple organs. It can appear with different clinical manifestations and nonspecific symptoms. Although local abscess formations secondary to brucellosis in different organs have been reported, no evidence for parotid involvement has yet been reported. In this study, a case with primary Sjögren's syndrome and parotid abscess secondary to brucellosis is presented.

Keywords: Sjögren's syndrome, parotid, brucellosis

Introduction

Sjögren's syndrome (SjS) is a chronic autoimmune disease affecting exocrine glands. Dryness of the mouth and eyes are the most common symptoms. However, patients with SjS may present with general systemic symptoms such as fever, malaise, fatigue, and weight loss (1, 2). Swelling of parotid salivary glands is observed in approximately one third of patients, and it is usually bilateral. This swelling is often episodic and regresses within a few weeks. In some cases, it may persist for a longer time (2). Patients with parotid gland swelling should be assessed in terms of malignancies, infections, amyloidosis, sarcoidosis, and SjS. Unilateral parotid swelling, especially if accompanied by severe pain and redness, should be monitored for suppurative bacterial infections (3-5).

Brucellosis is a zoonosis caused by bacteria belonging to the genus *Brucella*. It can be transmitted to humans via meat, milk, and urine of animals such as sheep, goats, cattle, buffalo, and pigs. It is characterized by an increasing degree of fever with chills and musculoskeletal complaints (6). Brucellosis is a systemic disease that may involve one or multiple organs. It can appear with different clinical manifestations and nonspecific symptoms. Its specific distinctive symptoms are not present so it can mimic many other diseases (6). Although local abscess formations secondary to brucellosis in different organs have been reported, to the best of our knowledge, no evidence for parotid involvement has yet been reported.

Case Presentation

A sixty-five-year-old female was admitted to our clinic with complaints of pain, swelling, and redness in the right side of her neck. She had hypertension for five years and ischemic heart disease for two years. She was regularly taking medications. She had dry mouth, dry eyes, joint pain, and recurrent painful redness and swelling in her neck for three years. Hard, painful, and inflamed swelling had developed in the submandibular region of the right side of the neck 20 days ago (Figure 1). She had a medical history of parotid swelling, appearing twice during the last three years.

Her physical examination revealed the following: temperature, 37.8°C; heart rate, 94/min; swelling, redness, and increased temperature in the right side of her neck; and tenderness in her wrists. Complete blood count revealed the following: WBC, 3000/mm³ (4000-10 000/mm³); hemoglobin, 11.9 g/dL (12.5-14.5 g/dL); and platelets, 231,000/mm³ (150,000-450,000/mm³). Erythrocyte sedimentation rate was 97 mm/h (<20 mm/h), and C-reactive protein (CRP) level was 4.47 mg/dL (<3.14 mg/dL). Routine biochemical parameters were within normal limits. HBsAg, anti-HCV, and anti-HIV tests were negative. RF titer was 121 IU/mL (<10 IU/mL), ANA (HEp2) was +++ positive with granular pattern, anti-La and anti-RO



Cite this article as: Yolbaş S, Bozgeyik Z, Artaş G, Koca SS. Parotid abscess secondary to brucellosis in a patient with primary Sjögren's syndrome. Eur J Rheumatol 2017; 4: DOI: 10.5152/eurjrheum.2017.16063.

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Submitted: 23.07.2016

Accepted: 5.03.2017

Available Online Date: 04.07.2017

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Figure 1. Incision performed for the swelling and drainage in the right side of the neck

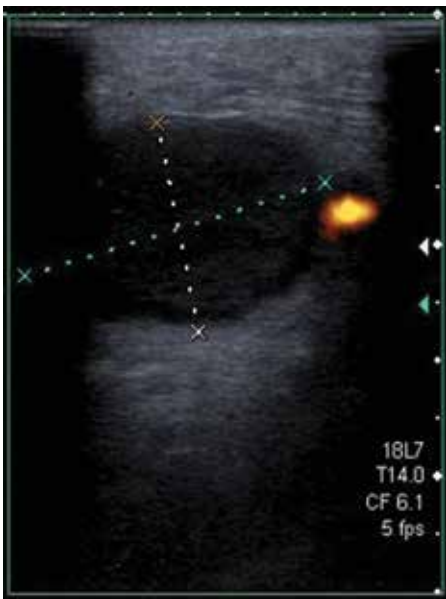


Figure 2. Neck ultrasonography; abscess formation in the smooth contour with internal echoes in the right parotid location

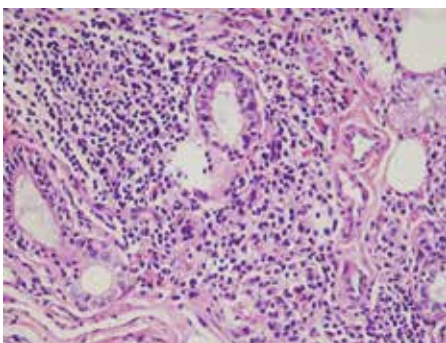


Figure 3. Histopathological examination of the minor salivary gland biopsy; periductal and perivascular lymphocytic infiltration and sometimes hyperplasia in the ductal epithelial cells were observed

tiers were >200 U/mL (<10 IU/mL). Although, *Brucella* standard tube agglutination test was negative, Coombs test was positive. On ultrasonography, a hypoechoic nonvascular and regular contoured mass (with 27×19 mm) which diameter was observed at the location of the right parotid gland was accepted as an abscess (Figure 2). Moreover, lymphadenopathy (2.5×1.5 cm) having hilar echoe was observed near this abscess.

On analysis of the abscess drainage fluid, acid-resistant basil staining showed negative results, no growth was noted on mycobacterial culture, and serological tests for tularemia were negative. Intravenous (IV) ceftriaxone 2×1 g, metronidazole 4×500 mg per oral (PO), and meloxicam 1×15 mg PO were empirically started. The patient did not have fever, and regression was observed in other signs and symptoms after this treatment. *B. melitensis* growth was noted on culturing the drainage fluid. She had a history of fresh goat cheese consumption. No other organ showed brucellosis. Anti-biotherapy was switched to doxycycline 200 mg/day PO and rifampicin 600 mg/day PO.

Schirmer test results were 2 and 5 mm on the right and left eyes, respectively. Tear break-up time was 5 s on the right and 6 s on the left. Minor salivary gland biopsy is performed in case of patients with a diagnosis of SjS. In histopathological examination, periductal and perivascular lymphocytic infiltration and hyperplasia in the ductal epithelial cells were observed, and this was found consistent with grade 3 Chisholm (Figure 3). She met the American College of Rheumatology (ACR)-2012 SjS classification criteria, and hydroxychloroquine 2×200 mg PO and artificial tears 7 times/day were added to the treatment regimen (7).

On the 7th day of treatment, swelling recovered. Brucellosis treatment was completed in 45 days. There was no recurrence after treatment for 6 months. The patient is still under our monitoring. However, leukopenia was ongoing, although brucellosis recovered. Thus, it is possible that leukopenia is related with SjS instead of brucellosis.

Discussion

Sjögren's syndrome is a chronic autoimmune disease characterized by mononuclear cell infiltration and progressive injury in exocrine glands. Typical findings of SjS are mouth and eye dryness. It can also cause a variety of symptoms by affecting exocrine glands in many other organs and systems. SjS is observed in 1-3% of the general population, and it often affects

women aged 40-50 years (1). Parotid glands are often affected in SjS. Expansion and swelling of the parotid gland showing intermittent course and often emerging as bilateral can be seen in patients as a result of ductal obstruction developing due to inflammatory process of the disease and secondary infections (4). The incidence of lymphoma in SjS is approximately 5% (2). Occurrence of swelling in the parotid gland, palpable purpura, low C4 complement level, presence of lymphadenopathy, and cryoglobulinemia increase the risk of malignant lymphomas in SjS (2).

While unilateral parotid growth is often seen in the presence of bacterial infections, chronic sialadenitis, duct obstruction, and neoplastic diseases (such as lymphoma and Warthin's tumor), bilateral growth is expected in the case of masseter muscle hypertrophy, viral infections (such as mumps and HIV), SjS, amyloidosis, sarcoidosis, acromegaly, anorexia, and alcoholism (3-5). Acute suppurative parotitis often leads to unilateral, sudden severe pain, swelling, and redness in the parotid location. It generally develops in case of excess dehydration, systemic diseases, and immunocompromised conditions. It predisposes oral bacteria to progress through the ducts, resulting in the development of secondary infections as a result of decreased ductal lavage, and it is mostly caused by streptococcal, staphylococcal, and anaerobic bacteria those are also a part of the oral flora (4).

In our patient, *B. melitensis* was observed in the culture of mai which was drained from parotid gland abscess. Brucellosis is a zoonosis primarily and often infects people who consume milk and other dairy products. It causes infertility in healthy animals. There are no typical signs of this disease in humans, but it is often characterized by symptoms such as fever, night sweats, fatigue, loss of appetite, and joint pain (6). Although it appears as undulant fever in the majority of patients, it can be less frequently seen as a local infection. Mild painful lymphadenopathy can be detected in the cervical and axillary regions by approximately 20% during the disease course. Several serological tests such as the standard tube agglutination, Rose Bengal, ELISA, and immunofluorescence tests are used in the diagnosis of brucellosis. Tube agglutination test is the standard method. The production of brucella in the culture from blood, bone marrow, or infected tissues is more specific diagnostic tool. Tetracycline, rifampin, streptomycin, trimethoprim/sulfamethoxazole, and third-generation cephalosporins can be used for treatment, depending on organ involvement (6).

Suppurative parotitis and abscess formation in the parotid gland may develop in the presence of certain predisposing factors (8). In addition, we did not detect secondary parotid abscess induced by *Brucella spp.*, although abscess formation related to *Brucella* in different parts of the body have been reported in the literature (9, 10). Repetitive intermittent swelling of the parotid gland is expected in SjS; however, patients must be evaluated for bacterial suppurative parotitis and abscesses due to decreased ductal flow, especially in cases of unilateral swelling and redness.

In conclusion, parotid abscess secondary to *Brucella* should be considered in the differential diagnosis of SjS parotitis, particularly where brucellosis is endemic, as brucellosis may present itself almost anywhere in the body with focal abscesses.

Ethics Committee Approval: N/A.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - S.Y., S.S.K.; Design - S.Y., G.A.; Supervision - Z.B., S.S.K.; Resources - S.Y.,

S.S.K.; Materials - Z.B., G.A.; Data Collection and/or Processing - Z.B., G.A.; Analysis and/or Interpretation - S.Y., S.S.K.; Literature Search - S.Y., S.S.K.; Writing Manuscript - S.Y., S.S.K.; Critical Review - Z.B., G.A.; Other - S.S.K.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

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