Chest Wall Involvement as a Manifestation of Brucellosis

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ABSTRACT

Brucellosis continues to be a common infectious disease in some parts of the world. Although the disease has different presentations, but chest wall involvement, as a manifestation of brucellosis is rare. In this study, we report three cases of chest wall involvement as manifesting feature of Brucellosis in Iran. They presented with a history of parasternal masses revealed to a diagnosis of Brucellosis and responded well to the treatment. Brucellosis may present with strange and unpredictable manifestations and can be misdiagnosed with tuberculosis and malignancies, especially in endemic areas for both TB and brucellosis.

Key words: Brucellosis, Chest wall involvement, Manifestation

INTRODUCTION

Brucellosis is primarily a disease of wild and domesticated animals, transmitted to humans through three major roots: consumption of unpasteurized dairy products, contact with infected animals, or environmental exposure.[1]

This multisystem and potentially lethal disease of zoonotic origin presents with variable, nonspecific clinical manifestations. It is endemic in the Mediterranean area, Middle East, western Asia, Africa, and Latin America. The true incidence is unknown and varies from about 0.01 to more than 200 per 100,000 population.[2,3]

Misdiagnosis of Brucellosis occurs because of these variable clinical pictures. Therefore, besides to the history of occupation (butchers, veterinarians, farmers, and employees of dairy industries) and travel, one should bear in mind that suspicion of Brucella infection is the cornerstone for correct diagnosis, especially in endemic regions.[1,3,4]

Localized disease may occur in many organ systems including the skeleton, urinary tract, central nervous system, liver, heart, and lung.[2,3]

CASE REPORTS

Case 1

A 23-year-old Sistani (a certain ethnicity resided in Sistan Province of Iran) shepherd admitted to the infectious disease ward due to 2 weeks intermittent low-grade fever (38°C), severe pain in left sternoclavicular joint (SCJ), progressive dyspnea, and gradual loss of consciousness. A fluctuant, soft and tender abscess (4 × 4 cm²) was found in his right SCJ area.

A thick wall hypoechoic mass was reported in ultrasonography. Bone appearance was normal in his chest X-ray (CXR) and computed topography scan (CTS). Brucella seroagglutination tests, Wright, and 2ME were requested with regards to his occupational state.

Partial sternotomy and necrotic debridement were done by a thorax surgeon and repaired by the surrounding muscles and soft tissues. Significant laboratory data were as followings: Erythrocyte sedimentation rate (ESR) = 65 mm/first hour, white blood cells (WBC) = 6800 (63% poly morpho nuclear), Wright = 1/2560, 2ME = 1/1280 and hemoglobin (Hb) = 9.2 g/dL.
Castaneda media culture of purulent material yielded to brucella melitensis infection. Smear and culture were negative for mycobacteria.

Cap. Doxycycline (100 mg/BID/Po), and Amp. Streptomycin (1 g/day, IM) were prescribed for 2 weeks and continued with Doxycycline (100 mg/BID) and Tab. Rifampin (600 mg/day) for 6 weeks. Complete recovery with no complication was reported after 6-month follow-up.

Case 2

An 18-year-old girl presented with a progressive bulging mass \(2 \times 3 \text{ cm}^2\) in size, mildly tender, and soft without any fluctuation in 3 weeks located in her rib inter-space 1-2 near the left border of the sternum. She was living in the urban area and was healthy otherwise, except for a history of unpasteurized cheese usage. Intermittent low-grade fever (at night) and sweating were the only complaints. CXR was normal and ultrasonography reported the mass as a lymphadenopathy. Brucella seroagglutination tests, Wright, and 2ME were requested.

Laboratory data showed: WBC = 11,200: Neut = 78%, Lymph = 20%, Mono = 2%, Hb = 11.1, Wright = 1/640, 2ME = 1/80, ESR = 9. Other lab data were not remarkable.

Biopsy of the lesion showed nonspecific inflammation of the lymph node. She was treated with Cap. Doxycycline (100 mg/BID/Po) and Tab. Rifampin (600 mg/day/Po). After 2 months, lymphadenopathy disappeared and no relapse was reported after 1 year follow-up.

Case 3

The third case was an 18-month-boy referred with a history of multiple, firm, and tender parasternal nodules (about \(1.5 \times 2 \text{ cm}^2\) in size). He showed no constitutional symptoms. There was no clear history of using unpasteurized dairy products. Wright and Coomb’s Wright and all workups were negative. Routine laboratory exams revealed normal results. As a diagnostic approach, excisional biopsy was performed. Grossly, the lesion was cartilaginous with no evidence of necrosis or tumoral involvement. Microscopic examination revealed chronic nonspecific inflammatory process. The smear and culture of biopsied material were negative for tuberculosis or other bacteria. Due to persistent signs and symptoms, follow-up was done by repeated serologic tests which showed fourfold rising in brucella titer (8 weeks later). Treatment was started by Tab. Trimethoprim–sulfamethoxazole (trimethoprim, 8 mg/kg/Po) and Tab. Rifampin (10 mg/kg/Po) and continued for 8 weeks. Good response to therapy was achieved.

**DISCUSSION**

Various clinical manifestations and different focal complications are the main problems for the correct diagnosis in Brucellosis.\[7\]

Blood, bone marrow, or tissue cultures are most often positive during the acute phase. In suspected brucellosis cases, prolonged culture for 21 days remains a mainstay approach.\[8\] In the absence of positive cultures, serologic tests could be helpful.\[8\]

The disease commonly presents as a febrile illness, and involvement of soft tissue is unusual in human.\[6\]

In a study in Turkey, focal complications were studied in 151 patients with brucellosis. Musculoskeletal complications were seen in 33.7% (30 spondylodiscitis, 15 sacroileitis, 5 peripheral arthritis, and 1 tendonitis), peritonitis and skin involvement were reported in 0.6%.\[9\]

Uncommonly, hematogenous spread of the disease may present as local infections in different organs.\[6,10-14\] Chest wall involvement (for example, cold abscess, rib involvement) in tuberculosis is a typical manifestation but in brucellosis is very unusual.\[6\] We did not find chest wall involvement (abscess, mass, etc) as the manifesting feature of brucellosis in the literature.

In our cases, smears were of no benefit because of nonspecific findings. Culture was positive in one case, surprisingly brucella melitensis was grown in pus culture from chest wall abscess, but serology was diagnostic in two other cases (although with delay in Case 3). In a study on 495 cases of brucellosis from India, in the absence of serologic tests, 88.7% of the patients had missed diagnosis.\[7\]

Despite years of research, the optimum therapeutic approach to brucellosis is not known well.\[2\] The recommendations of World Health Organization (WHO) expert on brucellosis are Tab. Rifampicin 600–900 mg and Cap. Doxycycline 200 mg daily for a minimum of 45 days in adults and in children, Tab. Rifampin should be substituted, with trimethoprim or sulfonamide as alternatives. Relapses can occur within 3–6 months of stopping therapy.\[9\] In our cases, no relapse was reported during follow-up.
CONCLUSION

It can be concluded that Brucellosis should be considered in the differential diagnosis of chest wall lesions especially in endemic areas. Repeated serologic tests for brucellosis could be useful in some unusual cases.

Finally, control of the disease in the animal reservoir, occupational precautions, and disinfection of dairy products, and other potentially contaminated products are among the methods for control of human brucellosis.\(^2\)

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