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Case Report

A Rare Case of a Brucellar Granulomatosis Mimicking Soft Tissue Tumor

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Abstract

Osteoarticular involvement is one of the most frequent complication of brucellosis. Upper extremity muscular involvement is an extremely rare manifestation of musculoskeletal brucellosis. Our case is a 60-year-old male patient who had presented with pain, swelling and limited range of motion of the left shoulder. Physical examination revealed two semi-mobile, soft-tempered mass in the anterior part of the left shoulder. After surgical procedure, cyst were excised and patological results had shown a granulomatosis lesion. Brucella tube agglutination test was found to be 1/320 positive. Purified protein derivative (PPD) was negative. *Brusellosis* can be located in different systems but upper extremity muscular involvement is rare. Brucellosis should be kept in mind especially in the endemic areas of brucellosis, in intramuscular abscesses of unknown etiology.

Keywords: *Brucellosis; Granulomatosis; İntramuscular Abscess; Surgery; Upper Extremity*

Introduction

Brucellosis is a zoonotic disease which can transmit to humans under certain conditions. Brucella species (spp)., which is the causative agent of the disease, is an intracellular, gram negative, oxidase positive, encapsulated and immobile rod. The main transmission pathway in brucellosis is milk products such as infected milk, cheese, cream and oil consumed without pasteurization [1]. Brucellosis can be presented with different kinds of clinic symptoms and can mimick other diseases.

Osteoarticular presentation rate of brucellosis is about 10 - 85% and the most common presentations are arthritis, bursitis, osteomyelitis [2]. The most common muscular involvements are psoas and paraspinal muscles [3]. In this article, we want to present a brucellosis case with atypical muscular and articular involvement.

Case Report

A 60-year-old male patient presented to the Orthopedics and Traumatology Outpatient Clinic with pain, swelling and limited range of motion of the left shoulder. The patient had no history of trauma. Two years ago, he had a swelling on his left shoulder and his pain had increased steadily over the last 6 months. The patient, who described the increasing pain and movement limitation in the arm with increasing swelling, had received analgesic and amoxicillin clavunate 2 x 1000g treatment for 2 weeks due to these complaints. The patient didn't respond to these treatments and presented to our hospital with the same complaints.

The patient's detailed history revealed no additional features. There was no pain, fever, weight loss, night sweats and other joints. The patient, who did not have a medical history, was dealing with livestock.

Physical examination revealed a 5 x 4 x 3 cm semi-mobile, soft-tempered mass in the anterior part of the right shoulder. At the posterior surface of the right shoulder, a 4 x 2 x 2 cm mass with the same features was found. The other systemic examination was normal (Figure 1).

Figure 1: *Clinical apperances of the patient*

The hemogram and biochemistry parameters of the patient were normal. Sedimentation rate was 71 mm/h and CRP was 10.7 mg/dL. Lymphocyte dominance was seen in peripheral smear. Magnetic resonance imaging of the patient revealed a mass lesion located in the thick-walled intramuscular space and the joint space showing intense enhancement followed by IVCM extending from the arm level at the inferior and extending to the supine and extending to the glenohumeral joint space. Humerus head was interpreted as deformed (Figure 2).

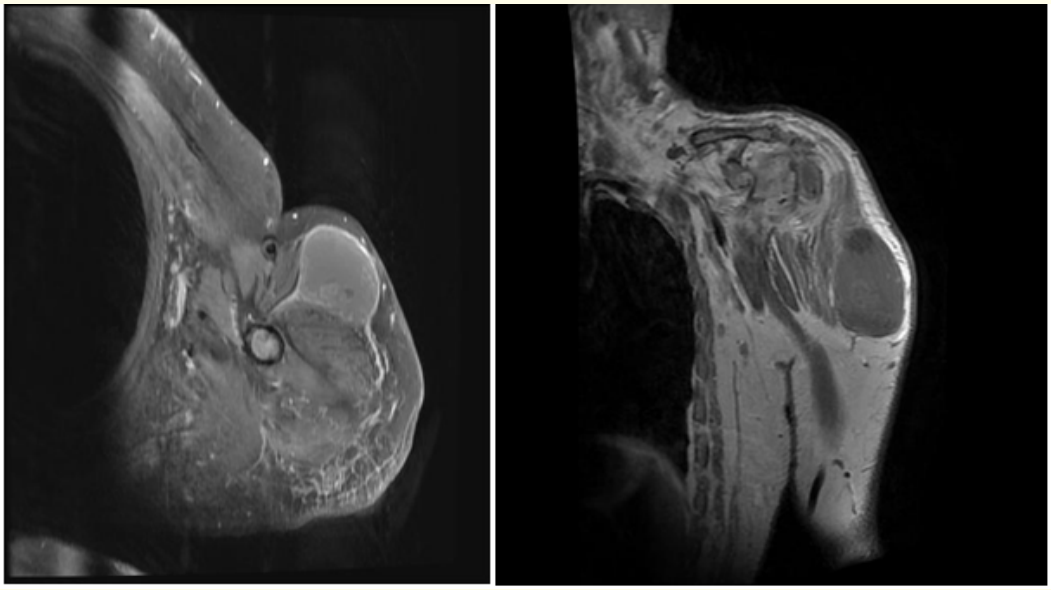

Figure 2: *MRI images of shoulder.*

The patient underwent operation for histopathological diagnosis with primary malignant soft tissue mass, sarcoma and metastasis, and a 10 cm incision was made on the left shoulder deltopectoral subcutaneous swelling. The cyst wall was bluntly dissected from the surrounding soft tissues and the muscles. Bright white colored lesions were present in the cyst with bleeding focus (Figure 3).

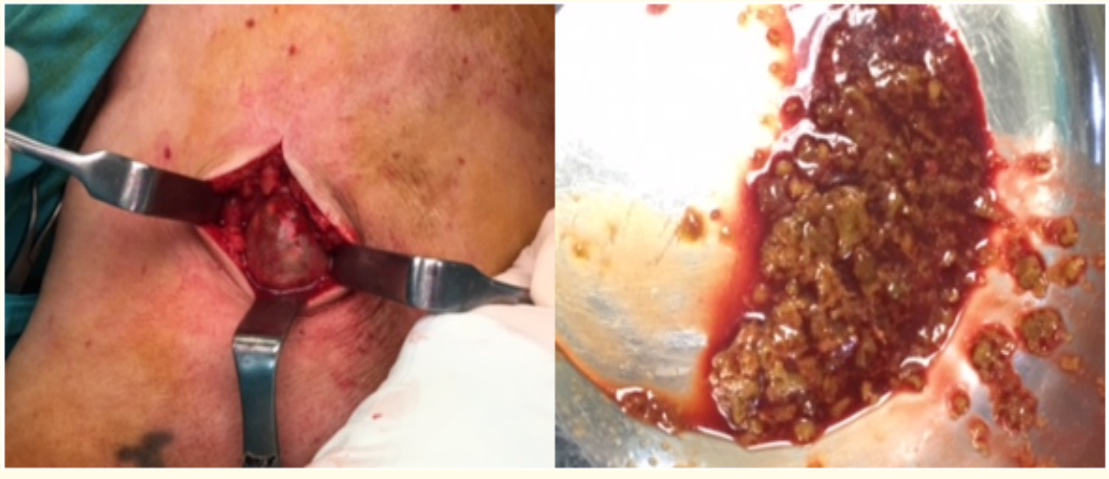

Figure 3: *Granulomatosis and bleeding lesions.*

The same procedure was performed in the lesion on the shoulder posterior. Cyst contents were sent to microbiological and pathological examinations.

The pathological examination was interpreted as compatible with granulomatous inflammation and in the sections granuloma structures were observed in places with excavation material and calcification areas (Figure 4).

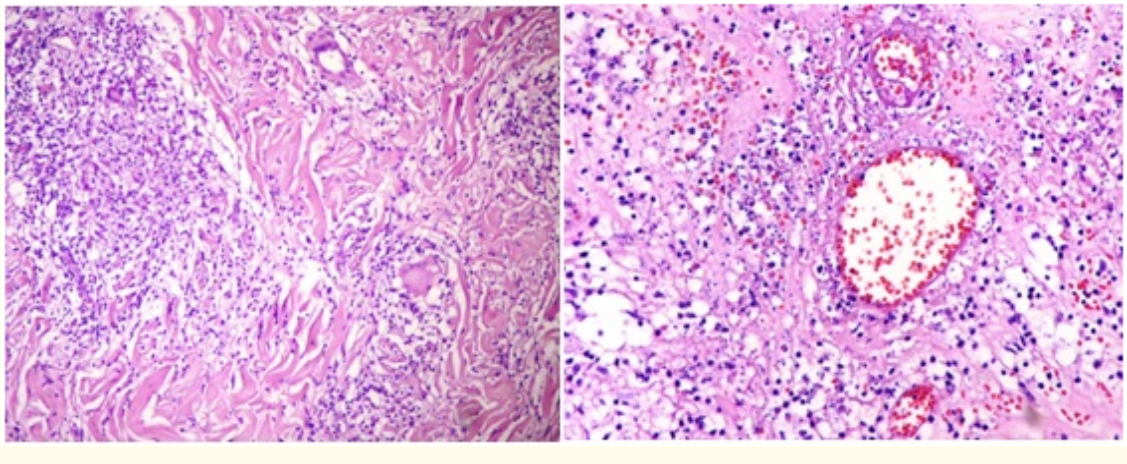

Figure 4: *Histopathological examination of the lesion.*

No microbiological growth was observed in nonspecific culture. Mycobacterium growth was not observed in the culture of tuberculosis. Brucella tube agglutination test was performed and tuberculin skin test (PPD) was performed for differential diagnosis. Brucella

tube agglutination test was found to be 1/320 positive. PPD was negative. The patient was treated with brucellosis and rifampicin 600 mg po and doxycillin 200 mg po treatment was started. The patient's treatment was completed in 12 weeks. At the end of the treatment, the sedimentation rate and CRP levels were in the normal range and there were no active complaints.

Discussion

Subcutaneous upper extremity lesions are generally originated from connective tissue, vascular, neural and bone masses. Connective tissue originated masses are ganglion cysts, giant cell tumors of tendon sheats, lipomas and fibromas. Vasculary lesions are hemangiomas. Bone derived tumors are enchondroma, osteochondroma and aneurysmal bone cysts. Other lesions are infective diseases or abscess, hematomas, myositis and myositis ossificans [4].

Brucellosis is a systemic disease that can attack every organ and system in the human body. Osteoarticular involvement rate of *Brucella* was reported to be 44 -7 6% in several publications [5,6] and the most common involving organ is paraspinal muscles and sacroiliac joint [3,7].

Brucellosis is an infective disease of world-wide distribution. It used to be endemic in the Mediterranean area, where it was first diagnosed and where it received its initial name 'Malta fever'. It is also known as 'undulant fever' or 'Mediterranean fever' or 'Rock Gibraltar fever'. The involvements of bones and joints may adopt many forms: Articular pains, Acute arthritis, Osteitis, Chronic arthritis. Brucellar spondylitis has been described in this last category. Papathanassiou., *et al*. reported [8] small number of our series (six patients) shows the low incidence of brucellar spondylitis. The lumbar spine is an elective localisation. The treatment of Brucellar spondylitis is conservative, as opposed to the surgical management of other complications with bone and joint involvement. All patients showed no signs of recurrence.

In our case, the involving region was the left shoulder. In our literature review, upper extremity involvement was rare. Kojan., *et al.* [9] reported a 16 years old myositis in deltoid muscle. Batmaz., *et al*. [10] reported a case about spondilodiscitis and dactilitis involvement in a same patient. In the meantime Turan., *et al*. reported [3] a case involvement of iliacus muscle, olecranon bursitis. In our cases we have no other joint or region involvement and disease only affected shoulder joint and intramusculary abscess.

Conclusion

Brucellosis is a systemic zoonotic infectious disease, which may present with different clinical presentations that may affect the organs and systems [10]. This disease, which is endemic in many Mediterranean countries like Turkey, is a serious public health problem [11]. Brusellosis can be located in different systems but upper extremity muscular involvement is rare [12]. Brucellosis should be kept in mind especially in the endemic areas of brucellosis, in intramuscular abscesses of unknown etiology.

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Volume 10 Issue 2 February 2019