Brucellosis as a primary cause of tenosynovitis of the extensor muscle of the arm

La brucellosi quale causa primaria di tenosinovite del muscolo estensore del braccio

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INTRODUCTION

Brucellosis is a kind of zoonosis that usually affects the musculoskeletal system. Musculoskeletal involvement of brucellosis varies between 10% and 85%. Osteoarticular involvement includes sacroiliitis, spondylitis, peripheral arthritis, bursitis, and tenosynovitis [1, 2]. Involvement of the hand and wrist is not common, and development of secondary tenosynovitis in the course of a brucellar infection is even much more unusual [3,4]. Brucellar tenosynovitis of the extensor tendon sheath is an extremely rare manifestation of musculoskeletal brucellosis. As far as we know, this is the first case to report the extensor tenosynovitis of primary brucellosis.

CASE REPORT

A 36-year-old male patient presented with described pain in his right wrist that started six days earlier. There was also a diffuse swelling in the 1st finger of his right hand. The patient described fever, night sweats, widespread muscle pain and fatigue that had been going on for the last six days. He did not have any history of trauma, psoriasis, diarrhea, urethritis or any familial inflammatory arthropathy. Vital signs were normal during physical examination. On physical examination, his temperature was 38.2°C; pulse was 86 beats/min; respiration count was 18 min; and blood pressure was 110/70 mmHg. Other systemic examinations were also normal. He gave a history of unpasteurized dairy cheese consumption in daily life. Physical examination revealed neither splenomegaly, nor hepatomegaly. Lympadenopathy was also absent. The locomotor system examination revealed diffuse swelling and pain in the 1st finger of the right hand. There was mild swelling in the volar surface of the wrist, and we determined atrophies in thenar muscles and in the 1st and 2nd fingers, and limitation of flexor muscles in the right hand. White blood cell count was 9000 mm³, erythrocyte sedimentation rate was 33 mm/h and C-reactive protein was 12.1 mg/dL (normal range 0-5 mg/dL). The serum uric acid, calcium levels and urine test results were within the normal limits. The rheumatoid factor (RF), anti-nuclear antibody (ANA) and HLAB-27 tests were negative. However, the standard tube agglutination for brucella was positive at a titer of 1/320.

Magnetic resonance imaging (MRI) of the wrist showed minimal thickening in extensor carpi ra-
dialis tendon with an increased intensity, accompanied by the increase of local fluid around the extensor carpi radialis brevis and longus tendons (Figure 1). The diagnosis of brucellosis was based on the review of the patient’s history, clinical findings, and serological and radiological abnormalities. The treatment was started with a combination of doxycycline 200 mg/d, rifampicin 600 mg/d, and indometacin 100 mg/d. On the 12th day of the treatment, the swelling and pain in the wrist healed completely. Normal values of C-reactive protein and erythrocyte sedimentation rate were detected after treatment. Doxycycline 200 mg/d, rifampicin 600 mg/d and indometacin 100 mg/d were continued until the end of the 6th week, the limitation of the wrist movement and pain had improved progressively within the three weeks of the medical treatment.

**DISCUSSION**

Brucellosis is an infectious disease continuing to be a major health problem in Mediterranean countries [2]. It may involve all organs, while musculoskeletal system is more affected than others. Tenosynovitis depending by brucellosis occurs rarely [2]. Tenosynovitis is an inflammation of this sheath. The cause of the inflammation may be idiopathic, or it may result from diseases that cause inflammation, infection, injury, overuse or strain. The wrists, hands, and feet are commonly affected. However, the condition may occur in any tendon sheath [5]. Swelling, pain and movement restriction may be detected clinically in the affected area because of tenosynovitis. When clinical outcome gets worse, muscle atrophy and weakness develop. In present case, clinical manifestations progressed since pain, muscle atrophy and weakness existed. Symptoms were difficulty of the joint movement, swelling in the affected area, tenderness around a joint, especially in the hand, wrist, foot, or ankle as well as having pain during the movement of joint. Fever, swelling, and redness may indicate an infection, especially if a puncture or cut causes these symptoms. Laboratory findings were variable. Normal white cell count rate was 80%, although leukopenia, relative lymphocytosis, anemia, pancytopenia, and thrombocytopenia were described. The erythrocyte sedimentation rate increased up to 30% [3, 5]. In addition, our patient’s C-reactive protein and erythrocyte sedimentation rate were high. MRI is one of the methods for diagnosing osteoarticular complications of human brucellosis, especially during the early phase. Tenosynovitis is visible as fluid within the tendon sheath, with possible thickening of the sheath itself. Findings of synovial thickening and presence of fluid around the tendon sheath represent non-specific tenosynovitis. Using only the imaging methods alone may cause some difficulties for distinguishing inflammation from infective tenosynovitis. It
is important to define brucellar tenosynovitis because of the proper treatment to prevent complications. In this respect, MRI is an important imaging method to diagnose brucellar tenosynovitis, thickening of the tendons, and nerve compressions related to brucellosis, especially in the early phase of tenosynovitis [4-6].

There are a few previous reports for brucella tenosynovitis in the literature (Table 1). In these reports (2, 4, 5, 8), four brucellar tenosynovitis occurred on the hands and one occurred on the shoulder. In most of the cases good recovery after appropriate brucellosis treatment in endemic countries considered in diagnosis of brucella tenosynovitis (2, 4, 5, 8).

If the diagnosis of osteoarticular brucellosis is made earlier and is treated promptly, 90-95% of patients may reach their normal functions. In these cases, the goal of treatment is to relieve pain and reduce inflammation. Resting or keeping of the affected tendons still is essential for recovery. Furthermore, tenosynovitis caused by infection needs immediate treatment [5]. Essential treatment for brucellar tenosynovitis is a combination of medications for 6-8 weeks, with proper physical exercise of involved joints [9]. In some severe cases, surgery may be needed to release the pus around the tendon. If there is no infection, a steroid injection may be used to decrease inflammation along the tendon sheath. After recovery of tenosynovitis, strengthening exercises through using the muscles around the affected tendon may help to prevent the injury from recurring. The indication for surgical drainage includes history and physical examination consistent with acute or chronic tenosynovitis. In certain circumstances when acute tenosynovitis presents within the first 24 hours of infection, medical management may initially be used. Prompt improvement of symptoms and physical findings must achieve within the following 12 hours after the initiation of medical treatment; otherwise, surgical intervention will become necessary [5, 6, 9].

In conclusion, brucellosis is still an important infectious disease in Mediterranean countries. Tenosynovitis is an important feature of inflammatory process and it is an unusual complication of osteoarticular brucellosis. Brucellosis should always be considered by the specialists for the early diagnosis so that appropriate management of the disease can be initiated to prevent possible sequela in patients with tenosynovitis.

**Keywords:** Brucella, osteoarticular, tenosynovitis.
Il coinvolgimento osteoarticolare è la complicanza della brucellosi osservata con maggiore frequenza. La tenosinovite brucellare della guaina del tendine estensore è una manifestazione estremamente rara della brucellosi muscolo-scheletrica. Un paziente di 36 anni, maschio, è giunto alla nostra osservazione riferendo dolore al polso destro, con esordio sei giorni prima. Inoltre, si riscontrava un diffuso gonfiore al primo dito della mano destra. Il paziente riferiva anche febbre, sudorazione notturna, dolore muscolare diffuso e affaticamento manifestatisi negli ultimi sei giorni. Il test di agglutinazione in provetta per brucella risultava positivo, con un titolo di 1/320. Alla diagnosi, il paziente mostrava alterazioni radiografiche. Veniva dunque istituita terapia con doxiciclina 200 mg/die e rifampicina 600 mg/die per 6 settimane. Il trattamento medico ha consentito la completa risoluzione dell’infezione.