OLGU SUNUMU CASE REPORT

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Acute Monoarthritis of the Wrist due to Brucellosis: A Report of Two Cases

Bruselloza Bağlı El Bileğinin Akut Monoartriti: İki Olgu Sunumu

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ABSTRACT Brucellosis is one of the most common zoonotic diseases in the world and is a public health problem in endemic countries. Rarely, acute monoarticular involvement can be the only symptom of brucellosis. Acute monoarthritis is a rheumatologic medical emergency. It is recommended that a patient with acute monoarthritis should be considered to have septic arthritis until proven otherwise. Infectious monoarthritis rarely affects the wrist. Also in cases which brucellosis has acute monoarticular involvement, it is exceptional that the wrist joint is affected. The presented patients admitted with pain, swelling and warmth in their wrists. Both cases had a history of occupational contact with animals such as sheep and goat and lived in a rural area endemic for brucellosis. Brucellosis should be considered in the differential diagnosis of monoarthritis of the wrist in areas where brucellosis is endemic or in patients coming from these regions.

Keywords: Arthritis; brucellosis; wrist; pain; case report

ÖZET Bruselloz dünyadaki en yaygın zoonotik hastalıklardan biridir ve endemik ülkelerde bir halk sağlığı sorunudur. Nadiren akut monoartiküler tutulum brusellozun tek semptomu olabilir. Akut monoartrit, romatolojik bir tıbbi acil durumdur. Akut monoartrit aksi ispatlanana kadar septik artrit olarak kabul edilmelidir. Enfeksiyöz monoartrit el bileğini nadiren etkiler. Ayrıca brusellozun akut monoartiküler tutulum gösterdiği durumlarda el bileği ekleminin etkilenmesi istisnai bir durumdur. Sunulan olgular el bileğinde ağrı, şişlik ve sıcaklık şikâyeti ile başvurdu. Her iki olguda da koyun, keçi gibi hayvanlarla mesleki temas öyküsü vardı. Her iki olgu da bruselloz için endemik olan kırsal bir bölgede yaşıyordu. Brusellozun endemik olduğu bölgelerde veya bu bölgelerden gelen hastalarda el bileği monoartritlerinin ayırıcı tanısında bruselloz düşünülmelidir.

Anahtar Kelimeler: Artrit; bruselloz; el bileği; ağrı; olgu sunumu

Acute monoarthritis is a rheumatologic medical emergency.[1] Brucellosis, a cause of monoarthritis, is one of the most common zoonotic diseases in the world; however, it is rarely seen as a septic arthritis agent.[2] Brucellosis is a public health issue in high-risk countries and is a neglected disease worldwide. Facultative, intracellular *Brucella* species cause brucellosis. *Brucella* abortus, *Brucella* melitensis and *Brucella* suis are the most pathogenic for both humans and their target hosts.[3] It is a multisystem disease that can show a wide variety of clinical manifestations. Rarely, acute monoarticular involvement can be the only symptom of brucellosis. In cases which brucellosis has acute monoarticular involvement, it is exceptional that the wrist joint is affected.[4] Herein, two cases of brucellosis occurring as acute monoarthritis of the wrist in an endemic region were presented.

CASE REPORTS

CASE 1

A 34-year-old right-hand-dominant male was admitted to our department with pain and swelling on his left wrist. His complaints had been going on for 3 weeks. He had a history of hand trauma from arm wrestling. His wrist pain was progressive in severity, continuous, and associated with the inability to bear

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weight. One week after the symptoms started, the patient sought medical care in the emergency room (ER). In the ER, he was diagnosed with tendinitis and treated with non-steroid anti-inflammatory drugs (NSAID) and wrist rest with an elastic bandage. Two weeks later, he was referred to physical medicine and rehabilitation (PM&R) clinic because his complaints persisted. He stated that his pain was not relieved by the NSAID drugs. His medical history showed no other diseases. There was no history of his medication, weight loss, fever, night sweats, fatigue, or insomnia. His occupation was animal husbandry.

Physical examination revealed redness, warmth, tenderness, and swelling in the left wrist. His passive and active left-wrist flexion and extension were restricted because of pain and swelling. The metacarpophalangeal squeeze test was positive for the left side. The examination of her remaining musculoskeletal system and other body organs was unremarkable. Laboratory blood tests showed a normal leukocyte count of $4,880\times10^3/mm^3$. The differential count showed decreased lymphocytes (18%) and increased neutrophils (72.9%). An increased C-reactive protein (CRP) level (35.5) was also noted. His liver and renal parameters were within normal limits and his rheumatoid factor was negative. Brucellosis was suspected due to the patient's complaints, physical examination findings, elevation in acute phase reactants, and occupational animal contact history.

CASE 2

A 29-year-old right-hand-dominant female with known diabetes presented to PM&R clinic with non-traumatic pain and swelling on her left wrist since 3 days. Her pain was constant, increasing at night and prevented her from using her hand. There were no other systemic symptoms. Her occupation was farming and animal husbandry.

Her left wrist was swollen and warm. Active and passive joint movement of her wrist was painful. Hemogram, liver and kidney function tests showed no abnormality. Acute phase reactants were within normal limits. *Brucella* was suspected due to her physical examination findings and occupation.

The Rose Bengal Test for brucellosis serological diagnosis was positive in both cases. Anteroposterior and lateral wrist X-rays of the cases showed soft tissue swelling around the left wrists. In Case 2, ultrasonography detected radiocarpal joint effusion (Figure 1). The patients were referred to the infectious diseases and clinical microbiology department in a hospital where the titration of the coombs serum test could be performed. The titration of the coombs serum was 1/360 in Case 1 and 1/5,120 in Case 2. The CRP of Case 2, who applied on the third day of her complaints, was found to be increased (28.1) in the second week of her complaints. The treatment regimen for brucellosis was initiated by doxycycline (100 mg orally every 24 h for 6 weeks), and rifampin (300 mg orally every 12 h for 6 weeks). Patients' condition improved dramatically after the start of the pharmacological treatment. No adverse events were observed.

Written informed consent forms were obtained from both patients to publish the demographic, clinical features and imaging findings. All procedures in these case reports were carried out in accordance with the latest version of the World Medical Association Declaration of Helsinki and the Good Clinical Practices Guidelines published by the Ministry of Health.

DISCUSSION

Brucellosis is a public health problem in endemic countries.[5] The presented cases are from a rural area where *Brucella* is endemic.[6] Peripheral arthritis in brucellosis is known to involve mostly large joints. In these cases, the wrist joint, which is rarely involved, was affected.[3,7] Although osteoarticular involvement of brucellosis is common, its manifestation as wrist monoarthritis is rare. Brucellosis should be kept in mind as an important cause of acute monoarthritis in emergency departments and units dealing with pain in endemic areas.

It is recommended that a patient with acute monoarticular arthritis should be considered to have septic arthritis until proven otherwise.[8] Having one or more hot swollen joints in a patient is a medical emergency and the most important cause of this condition is septic arthritis, which has serious morbidity and mortality.[2] The joint affected in 40-50% of in-

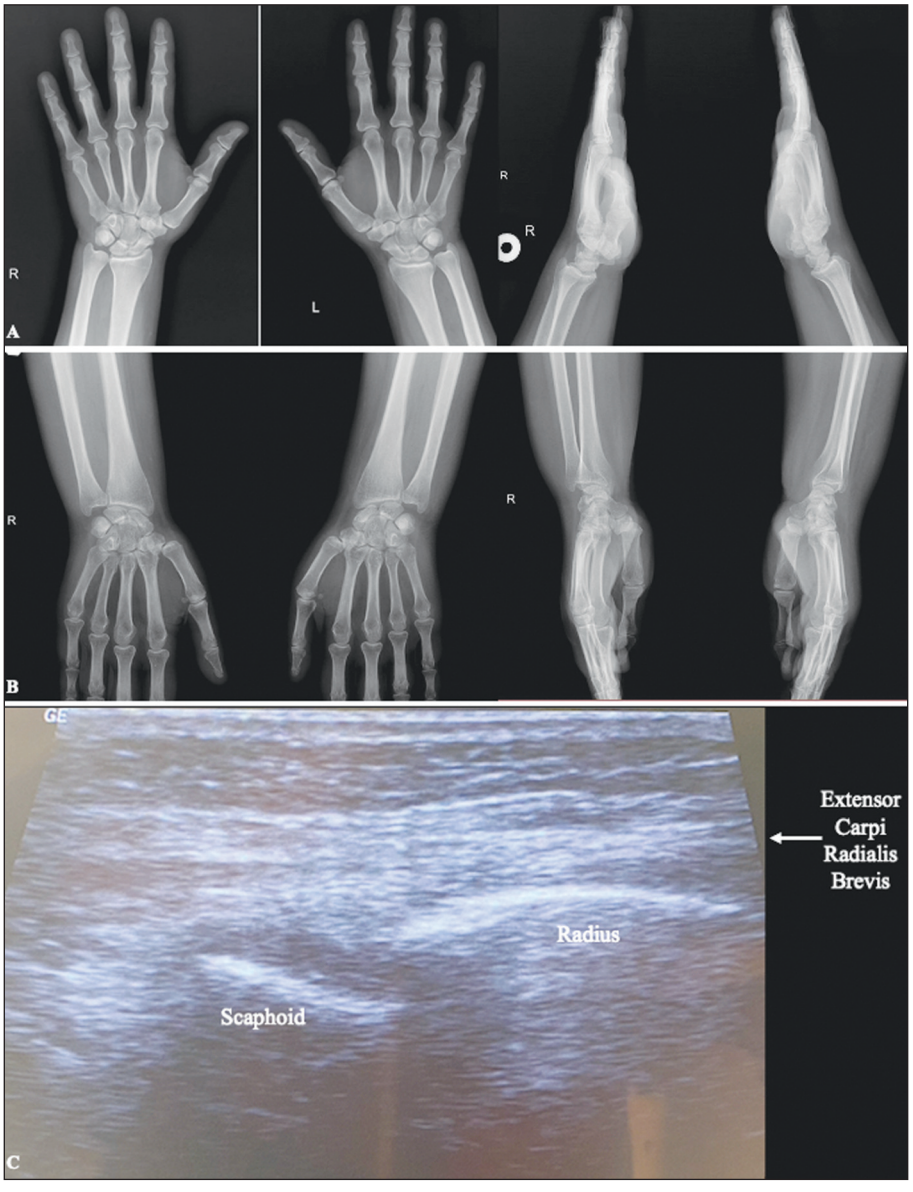

FIGURE 1: Wrist X-rays of Case 1 **(A)** and Case 2 **(B).** Ultrasound image of Case 2 **(C)** showing radiocarpal effusion. Note the anechoic (black) fluid just in front of the scaphoid.

fectious monoarthritis cases is the knee joint. This is followed by the hip in 13-20% of cases and the shoulder in 10-15%. Infectious monoarthritis rarely affects the wrist (5-8%).[8]

Patients with infectious wrist arthritis often present with a swollen, red, and painful joint and have a history of penetrating trauma. When the trauma is not obvious, the differential diagnosis includes degenerative arthritis, inflammatory arthritis, crystal arthropathy, cellulitis, and soft tissue abscess.[9] In these cases, there was no history of penetrating trauma or other conditions included in the differential diagnosis.

Rare organisms such as *Brucella* can also be detected as a causative agent of monoarthritis in endemic areas. The risk factors for brucellosis are ingestion of unpasteurized dairy products or occupational exposures.[8] The fact that the patients' occupation was animal husbandry, direct contact with sheep and goats, and consumption of raw milk products indicate that they had brucellosis risk factors.

Brucellosis is one of the most common zoonotic diseases all over the world.[4] *Brucella* spp is a small, aerobic, toxin-free, gram-negative coccobacillus that does not contain capsules, spores, or flagella. Bacterial growth is slow. They can avoid the protective activities of the organism by reproducing intracellularly. The organism is resistant to drying and freezing, sensitive to sunlight and heat. Various species of the genus *Brucella* prefer different hosts.[3,10] Humans are accidental hosts of brucellosis who acquire the disease through a variety of routes, including oral, conjunctival, respiratory, cutaneous, transplantational, blood and, rarely, bone marrow transplantation. Brucellosis typically occurs through direct exposure to contaminated animal products, genital secretions, aborted fetuses, infectious aerosols, and accidental vaccine inoculations.[3] *Brucella* melitensis (goat, sheep, cattle, camel) is the most virulent species for humans. *Brucella* suis (pig, cattle), *Brucella* abortus (horse, cattle), and *Brucella* canis (dog) are other common species. The bacteria can live for two months in cheese made from goat or sheep milk.[3,10]

Brucellosis progresses with multisystemic involvement.[4] One of the common complications of brucellosis is osteoarticular involvement with 10% to 85% of patients affected.[4]

Osteoarticular involvement in brucellosis can be seen at any stage of the disease and can be acute, subacute, or chronic. Although the most important clinical forms of osteoarticular brucellosis are arthritis, spondylitis, bursitis, tenosynovitis, and osteomyelitis, any region of the musculoskeletal system can be affected. Osteoarticular brucellosis is most commonly encountered as sacroiliitis or peripheral arthritis or spondylitis. Brucellosis affects the knee or hip joints more frequently than the peripheral joints. Involvement of costochondral, sternoclavicular, shoulder, elbow, wrist, metacarpophalangeal, ankle, and symphysis pubis joints is rare in brucellosis.[3,7,10] In these cases, acute monoarthritis of the wrist, a rare form of brucellosis, was detected.

Worldwide, approximately 500,000 new cases of brucellosis are reported annually, and it is endemic in the Mediterranean basin, the Middle East, Central Asia, China, the Indian subcontinent, sub-Saharan Africa, Mexico, and parts of Central and South America.[5] Türkiye is one of the countries where brucellosis is endemic. It is especially common in animals in the Ankara plain, Konya region, Diyarbakır and Şanlıurfa regions in Southeastern Anatolia. Brucellosis in humans is defined as an occupational disease often seen in butchers, slaughterhouse workers, veterinarians, animal breeders, farmers, and dairy workers.[6] The presented cases were detected in the Karapinar district of Konya province, which is a rural area in Türkiye.

The Centers for Disease Control recommends that unpasteurized dairy products and undercooked meat be avoided and that those handling animal tissues wear gloves, goggles, and aprons to prevent brucellosis.[11]

Serology, often combined with standard agglutination tests, is the basis of diagnosis in endemic areas. Blood cultures require prolonged incubation and have unpredictable sensitivity ranging from 53% to 90%. When the disease is limited to a single joint, blood cultures can be negative. Therefore serology is the basis of laboratory diagnosis. The synovial fluid culture can remain positive even though the blood culture is negative.[12]

Case 1 was incorrectly diagnosed with tendinitis in the ER due to a history of hand trauma from arm wrestling. *Brucella* was suspected in this case, as unresponsive to NSAIDs, history of animal contact, acute monoarthritis, admission from an endemic region, and physical examination findings. The anamnesis and the physical examination findings of Case 1, the elevation in the acute phase reactants, and the serological test positivity confirmed the preliminary diagnosis. The fact that Case 2 was a patient who admitted with acute monoarthritis from an endemic region and had animal contact due to her occupation suggested brucellosis. The laboratory findings of Case 2 were normal, but the anamnesis and physical examination findings, the positivity of the serological test, and the ultrasound findings confirmed the preliminary diagnosis. In addition, CRP had increased in the second week of her complaints.

Brucellosis typically presents as a nonspecific, flu-like illness characterized by fluctuating fever, headache, myalgia, arthralgia, lymphadenopathy, hepatomegaly, and splenomegaly.[3,13] The absence of systemic com-

plaints in the presented brucellosis cases is atypical. As the symptoms of brucellosis are atypical and complications are common, it may be misdiagnosed and therefore treatment may be delayed.[13]

In conclusion, brucellosis should be considered in the differential diagnosis of monoarthritis of the wrist in areas where brucellosis is endemic or in patients coming from these regions.

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