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CASE REPORT

# Rare Osteoarticular Involvement of Brucella Infection: Prepatellar Bursitis Presenting as a Massive Mass

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## Abstract

The osteoarticular involvement of Brucella infection may present as arthritis, bursitis, tenosynovitis, or spondylitis. Isolated presentation of Brucella infection as bursitis is a rare occurrence. The objective of this report was to present two cases of Brucella infection presenting with isolated prepatellar bursitis as osteoarticular involvement. The initial case report concerns a 56-year-old female patient who presented with a complaint of swelling in the left knee, accompanied by minimal pain, which had been present for the previous year. The second patient was a 53-year-old male patient who presented with a complaint of painless swelling in the knee, which had first appeared two years prior and had progressively worsened. Physical examination observed swelling extending from the anterior to the lateral side of the left knee, with no erythema, warmth, or tenderness observed in both patients. Laboratory examinations revealed normal biochemical parameters, complete blood count, and erythrocyte sedimentation rate (ESR), but C-reactive protein (CRP) levels were three times higher than the cut-off value. Serological tests revealed Brucella IgG positivity. Following the investigations, surgery was planned, and through a longitudinal incision, the masses were reached and completely excised. The resected tissue's pathological examinations revealed chronic granulomatous inflammation. In endemic regions such as the one under consideration here, Brucella bursitis should be considered in the context of isolated and suspected osteoarticular involvement, regardless of the presence of any features in the patient's history. Clinical suspicion plays a pivotal role in the preliminary diagnosis of Brucella bursitis, guiding clinicians in the early diagnosis and effective treatment of the disease.

**Keywords:** Brucella infection, osteoarticular involvement, prepatellar bursitis, knee pain, animal husbandry.

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## INTRODUCTION

Brucella infection is a zoonotic disease that can affect multiple organ systems, with the osteoarticular system being one of the most commonly affected (1,2). It is a significant public health problem worldwide (3,4), and the clinical presentation can range from mild to severe, involving multiple organ systems and mimicking other diseases. The initial diagnostic process can be complicated, and the disease's tendency to mimic other conditions further complicates this situation. The complexity of diagnosis and treatment necessitates a robust healthcare system, which is particularly important in cases where delayed diagnosis and treatment can impose a significant burden.

The osteoarticular involvement of Brucella infection may present as arthritis, bursitis, tenosynovitis, or spondylitis. Isolated presentation of Brucella as bursitis is a rare form of the disease (5,6). The objective of this report is twofold: firstly, to present two cases of Brucella infection presenting with isolated prepatellar bursitis as osteoarticular involvement and their treatment process; secondly, to conduct a review of the existing literature on this subject.

## CASE REPORT

### *Case-1*

The first case is that of a 56-year-old female who has been experiencing swelling in her left knee for the past year, accompanied by minimal pain. She has no occupational history associated with brucellosis, such as livestock farming, and no history of consuming unpasteurized milk or dairy products. With regard to comorbidities, she has no additional diseases except diabetes mellitus. Upon questioning her symptoms, she only reported swelling in the left knee and described minimal pain during joint movement. No additional symptoms such as fever, weight loss, headache, muscle or joint pain, fatigue, trauma, or night sweats were observed or reported. The patient had not taken any medication for the left knee swelling, but she reported undergoing three aspiration procedures at an external center, after which she noticed recurrent swelling.

Physical examination revealed swelling extending from the anterior to the lateral side of the left knee, with no erythema, warmth, or tenderness (Fig. 1). Joint movement was minimally limited in flexion and extension. Laboratory examination revealed normal biochemical parameters, complete blood count, and erythrocyte sedimentation rate (ESR), however, C-reactive protein (CRP) levels were three times higher than the cut-off value. Concurrent serological tests yielded a positive result for Brucella IgG, while the Rose Bengal test and the Brucella agglutination test (Coombs serum test) both returned positive results, with a titer of 1:1520.

Radiological examinations revealed a soft tissue mass extending from the anterior to the lateral side in both the coronal and sagittal planes, localized in the prepatellar region. Magnetic resonance imaging (MRI) revealed a thick-walled structure in the prepatellar area extending laterally toward the joint, measuring approximately 100×80×80 mm. The lesion contained hypo-intense foci on T2-weighted sequences, consistent with rice bodies, and a potential diagnosis of complicated prepatellar bursitis was made (Fig. 2).

Following the investigations, the surgical intervention was planned. A longitudinal incision was made extending from the superolateral aspect of the patella to the midline of the patella, and the mass was reached and completely excised (Fig. 3). The resected tissue's pathological examination revealed two cystic lesions, with chronic granulomatous inflammation, consistent with prepatellar bursitis.

Postoperatively, complete blood count, biochemical parameters, and sedimentation rate returned to normal. CRP levels decreased, and the patient reported no recurrence of symptoms. Follow-up examinations revealed no additional complaints. The patient was referred to the Infectious Diseases clinic in order to ascertain the multi-organ involvement of brucellosis. Screening tests revealed no other underlying conditions. Because of the confirmed diagnosis of Brucellosis, the patient was treated with rifampicin (600-900 mg/day oral) and doxycycline (200 mg/day oral) for six weeks as recommended in the literature (7). No complications and recurrence were encountered during follow-up.

### *Case-2*

A 53-year-old male patient presents with a complaint of painless swelling in the knee, which first appeared two years ago and has progressively worsened. His profession is animal husbandry, and he reports no fever, chills, headache, myalgia, fatigue, loss of appetite, weight loss, trauma or night sweats during this period. He has not used any medication related to his complaint, and be-

fore this period, he has not had similar complaints or symptoms. A physical examination revealed no signs of pain, tenderness, elevated skin temperature, or restricted knee movement. Laboratory tests revealed normal hemoglobin, white blood cell, and leukocyte values, as well as biochemical parameters, sedimentation rate, and C-reactive protein (CRP), with only a minimal increase in platelet count. Serological tests for brucellosis revealed increased levels of Brucella Ig G and IgM. Furthermore, the Rose Bengal Test and the Brucella Agglutination Test were observed as positive. A review of the radiological imaging revealed an extra-nodal mass extending laterally in the soft tissue in the coronal plane of the left knee and in the sagittal plane. MRI images showed an oval-shaped, well-contoured cyst with multiple septation within the prepatellar bursa, measuring approximately 5.6*9*10.6 cm (Fig. 4).

Following the conclusion of the examinations, the surgical procedure was initiated. The mass was completely resected through a prepatellar incision (Fig. 5). The resected tissues were analyzed, and the dimensions of the cyst were recorded as 9*9*1cm. The pathological findings were consistent with chronic synovitis. Post-surgery, no growth was observed in tissue culture and synovial fluid culture. Blood parameters remained stable, and the previous complaints were fully resolved. Consequently, screening tests performed in collaboration with the Infectious Diseases clinic did not reveal multi-organ involvement, and following the administration of rifampicin (600-900 mg/day oral) and doxycycline (200 mg/day oral)

treatment for six weeks (7), no recurrence, and further complaints were observed in the one-year follow-up.

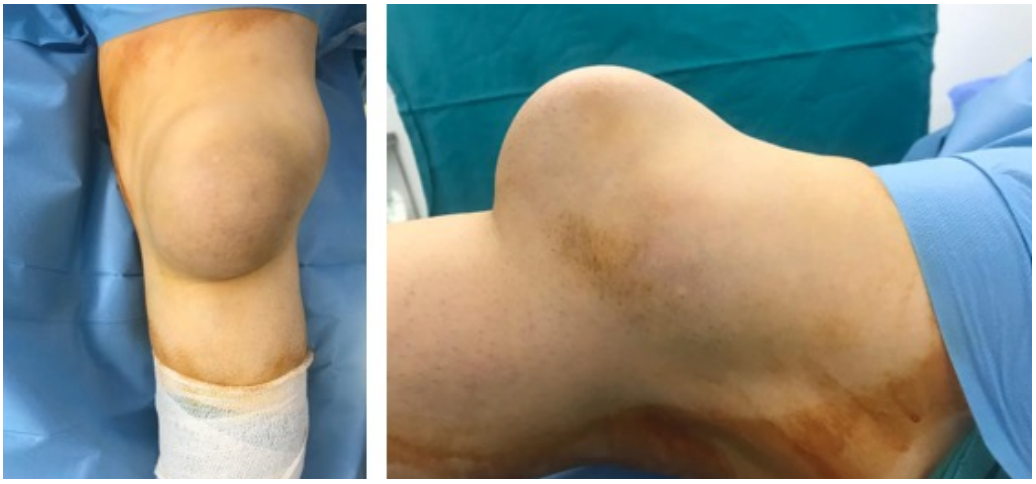

**Figure 1:** Flowchart of the study.

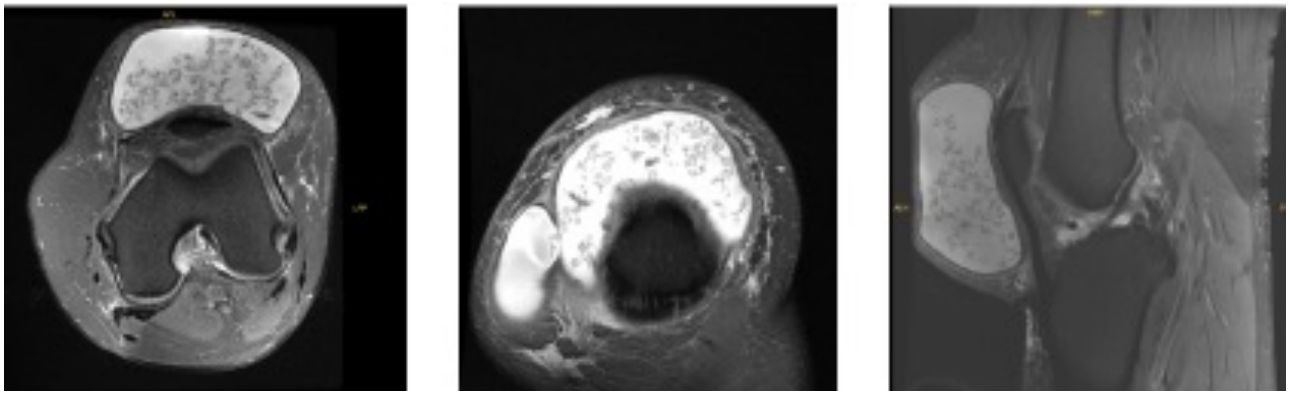

**Figure 2: F**irst patient's MRI images revealed hypo intense foci on T2-weighted sequences consistent with rice bodies, with a potential diagnosis of complicated prepatellar bursitis.

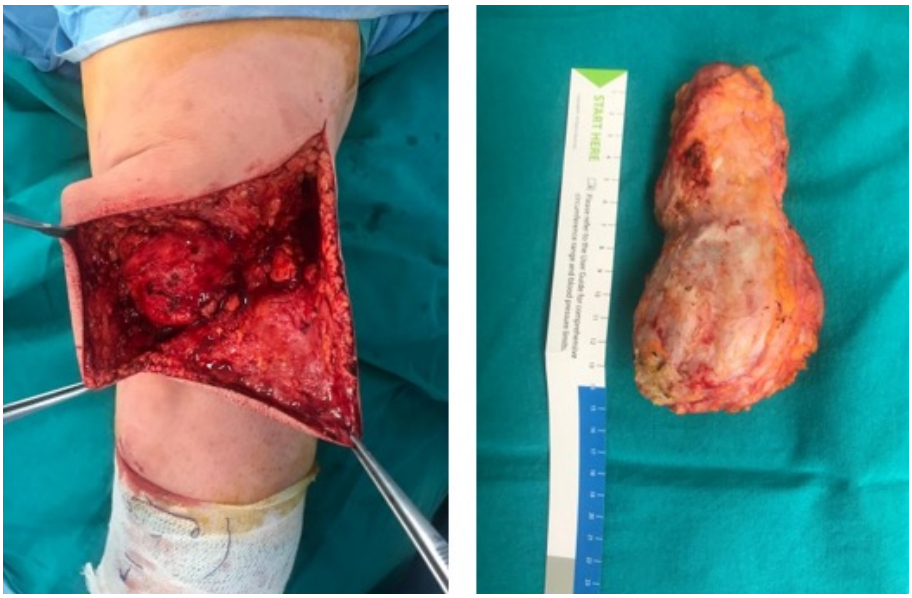

**Figure 3:** Through a longitudinal incision, the mass was completely excised

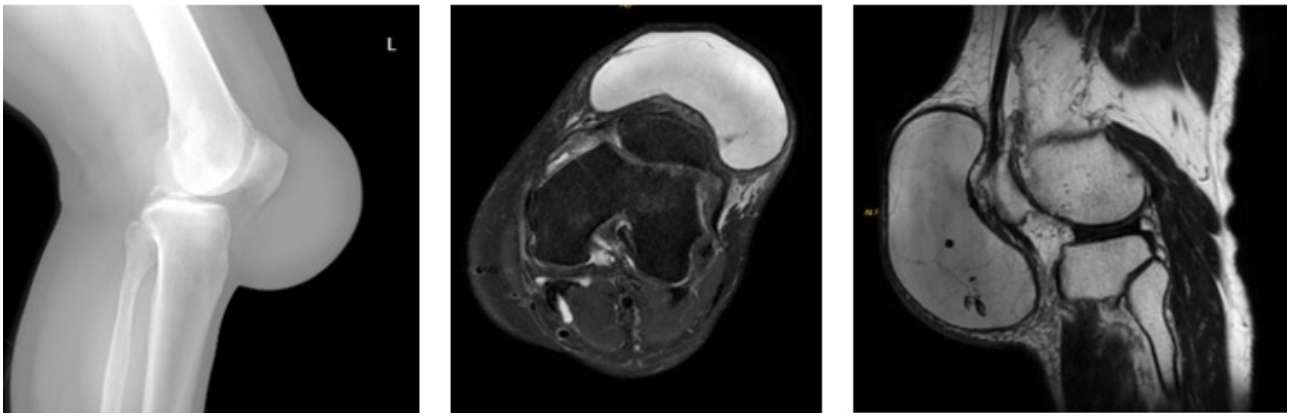

**Figure 4:** An oval shaped, well-contoured cyst with multiple septation within prepatellar bursa can be observed both on direct radiography and MRI images.

**Figure 5:** Through a prepatellar incision, the mass was completely resected

## DISCUSSION

Brucella bacteria are classified as gram-negative, facultative intracellular coccobacilli (1). Among the various species that cause brucellosis, Brucella melitensis is the most common cause of human brucellosis worldwide (8). The disease under discussion is eradicated in developed countries; however, it remains prevalent in developing nations. Brucella infection, an endemic disease, is most commonly observed in regions such as India, Mexico, Central and South America, the Mediterranean basin, and the Persian Gulf (9). Turkey is also among the endemic regions. The most significant aspect of this report is its detailed presentation of two different cases of osteoarticular involvement due to Brucella infection. In our country, where the disease is endemic, it is important to be prepared for isolated osteoarticular involvement of this nature. These two case examples highlight that, in cases of delayed treatment, surgical intervention becomes the primary treatment option, superseding medical therapy. It is imperative that medical professionals prioritize suspicion and clinical examination as key factors in diagnosis, as emphasized by our colleagues.

Brucella infection is a systemic disease that can affect multiple organ systems. Transmission occurs through contact with infected animals, consumption of unpasteurized milk from infected animals, or dairy products derived from such milk. One of the cases had a history of livestock exposure, while the other did not. The presentation of the infection can vary, ranging from acute to chronic forms. Acute cases typically present with nonspecific symptoms such as fever, night sweats, back pain, headache, arthralgia, myalgia, depression, and anorexia. In subacute cases, the condition may manifest as fever of unknown origin (10). The nonspecific nature of these symptoms poses a significant challenge to diagnosis. In both cases, the only observed symptoms were isolated swelling of the knee, with no other significant findings on physical examination. This underscores the pivotal role of clinical suspicion in the diagnosis of brucellosis.

In addition to nonspecific symptoms, Brucella infection can also present with localized organ involvement. The most common complication is osteoarticular involvement, which may occur as sacroileitis, spondylitis, peripheral arthritis, bursitis, osteomyelitis, or tendinitis (11-13). Beside these, infection of the bursa is rare and Brucella bursitis reported in only 1–7% of cases in bone and joint infections (3,6). Despite its rarity, the prepatellar bursa is the most commonly affected site in bursal infections caused by Brucella infection (6). Mousa et al. (14) reported 1.2% of 169 patients with brucellosis has a tenosynovitis and bursitis. Al-Majid et al. (15) described an olecranon bursitis case that was serologically negative but positive in blood and aspirate cultures. Taşova et al. (2) reported bursitis in 5 (5.7%) of 87 brucella cases with osteoarticular involvement. Pourbagher et al. (5) identified olecranon bursitis in only 3 (1.2%) of 251 cases. Traboulsi et al. (6) highlighted that the prepatellar bursa is the most frequently affected site in brucella-associated bursal infections. As demonstrated, isolated bursal infections without other organ involvement are extremely rare. In both our cases, Brucella infection presented with an isolated rare osteoarticular involvement, a giant mass. In endemic regions, Brucella infection should be considered the differential diagnoses in patients presenting with atypical osteoarticular involvements, whether with or without concomitant findings. It is crucial to remember the significant role of clinical suspicion in such cases.

The present study was conducted with the objective of demonstrating the significance of incorporating brucellosis into the differential diagnosis of patients exhibiting isolated bursitis, particularly within endemic regions. As documented in the extant literature (36,7,11-13), osteoarticular involvement due to Brucella infections may manifest as isolated bursitis, albeit in exceedingly rare instances. Furthermore, many findings have been reported in patients infected with Brucella, the most common being fever, which was reported in 60% of cases (7). Notably, in the present study, neither fever nor any other clinical indication was observed in the two patients. However, brucellosis infection was identified, and the sole discernible finding was isolated massive bursitis.

The manifestation of Brucella bursitis with non-specific symptoms and physical examination findings can result in misdiagnosis and unnecessary treatment, which may prolong the disease process and increase the risk of complications. This risk is particularly elevated in endemic regions.

In endemic areas such as our country, Brucella bursitis should be considered in the context of isolated and suspected osteoarticular involvement, regardless of the presence of any features in the anamnesis or physical examination. Clinical suspicion is therefore pivotal in the preliminary diagnosis of Brucella bursitis, guiding clinicians in the early diagnosis and effective treatment of the disease.

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**Abbreviations list**

ESR: erythrocyte sedimentation rate
CRP: C-reactive protein
MRI: Magnetic resonance imaging

**Ethics approval and consent to participate**

Ethics committee approval was not obtained since the present study was a case report. Written informed consent forms were obtained from both patients.

**Consent for publication**

Two patients whose data were presented in our study provided written and verbal consent for the presentation of their clinical data in the case report.

**Availability of data and materials**

The datasets generated and analyzed during the current study are not publicly available, but are available from the corresponding author on reasonable request.

**Competing interests**

The authors declare no conflicts of interest.

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**Authors' contributions**

Idea/Concept: TBY, BG, AÇ. Desıgn: TYB, MMA, AÇ. Control/Supervısıon BG, ÖD. Data Collectıon And/Or Processıng: TBY, BG, MMA, AÇ. Analysıs And/Or Interpretatıon: BG, MMA, AÇ. Lıterature Revıew: BG, MMA, AÇ, ÖD. Wrıtıng The Artıcle: TBY, MMA, AÇ. Crıtıcal Revıew: BG, ÖD. References And Fundıngs: BG. Materıals: ÖD. Other: ÖD.

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