Eklem Hastalıkları ve Cerrahisi
Joint Diseases and Related Surgery

Case Report / *Olgu Sunumu*

Eklem Hastalik Cerrahisi
2019;30(3):333-337
doi: 10.5606/ehc.2019.70215

Rare form of brucellosis, subacromial and subdeltoid bursitis: A case report and literature review

Brusellozisin nadir bir formu, subakromiyal ve subdeltoid bursit: Olgu sunumu ve literatür incelemesi

Bilgehan Çatal, MD.

Department of Orthopedics and Traumatology, Medipol University Medical School, İstanbul, Turkey

ABSTRACT

Brucellosis is a zoonosis seen all over the world and is still endemic in certain parts of the world. Brucellosis is a systemic infection which involves multiple organs and tissues. Although musculoskeletal system involvement is frequent in brucellosis, bursal involvement is seen rarely. In this article, we present a case of subacromial and subdeltoid brucellar bursitis with positive serology and aspiration culture. Patient achieved complete recovery with rifampicin and doxycycline treatment, without any evidence of relapse. A high clinical suspicion is required for the diagnosis of brucellar bursitis.

Keywords: Brucellosis, bursitis, subacromial, subdeltoid.

ÖZ

Brusellozis dünya çapında görülen bir zoonozdur ve halen dünyanın belirli bölgelerinde endemiktir. Brusellozis pek çok organ ve dokuyu tutan sistemik bir enfeksiyondur. Bruselloziste kas iskelet sistemi tutulumu sık olsa da bursal tutulum nadir görülmektedir. Bu yazıda, pozitif serolojisi ve aspirat kültürü olan bir subakromiyal ve subdeltoid brusella bursiti olgusu sunuldu. Hasta herhangi bir nüks belirtisi olmaksızın rifampisin ve doksisiklin tedavisi ile tamamen iyileşti. Brusella bursitinin tanısında yüksek klinik şüphe gereklidir.

Anahtar sözcükler: Brusellozis, bursit, subakromiyal, subdeltoid.

Brucellosis is a zoonotic infection, caused by facultative intracellular bacteria of the genus Brucella.[1] The disease is still an important public health problem throughout the world, but principally in the Arabian Peninsula, the Mediterranean region, the Indian subcontinent, Mexico, and parts of Central and South America.[2-4] The transmission of Brucella from infected animals to humans occurs either by mainly ingestion of unpasteurized contaminated animal products, particularly milk, cream, butter and fresh cheese or occupational contact to animals.[1,5,6]

The disease is characterized by fever, sweating, generalized malaise, chills, anorexia, arthralgia, back pain, lymphadenopathy or hepatosplenomegaly. It may result in complications involving multiple organs. The musculoskeletal system is one of the most commonly affected systems in brucellosis.[7] The frequency of osteoarticular complications is in the range of 10-85% in various studies.[8-10] Peripheral arthritis, spondylitis, and sacroiliitis are the most common manifestations of osteoarticular brucellosis.[7-11] In rare cases, extraarticular soft tissue involvement of osteoarticular brucellosis such as tenosynovitis and bursitis were also reported.[12,13]

CASE REPORT

A 68-year-old male patient presented with history of painless right shoulder swelling ongoing for about six months. Physical examination revealed only swelling of right subacromial-subdeltoid bursa and movement restriction of the right shoulder joint (Figure 1). The patient was afebrile and there were no

Received: June 20, 2019 Accepted: July 17, 2019 Published online: October 24, 2019

Correspondence: Bilgehan Çatal, MD. Medipol Üniversitesi Tıp Fakültesi Ortopedi ve Travmatoloji Anabilim Dalı, 34810 Beykoz, İstanbul, Turkey.
Tel: +90 507 - 623 80 40 e-mail: drbilgehancatal@yahoo.com

Citation:
Çatal B. Rare form of brucellosis, subacromial and subdeltoid bursitis: A case report and literature review. Eklem Hastalik Cerrahisi 2019;30(3):333-337.

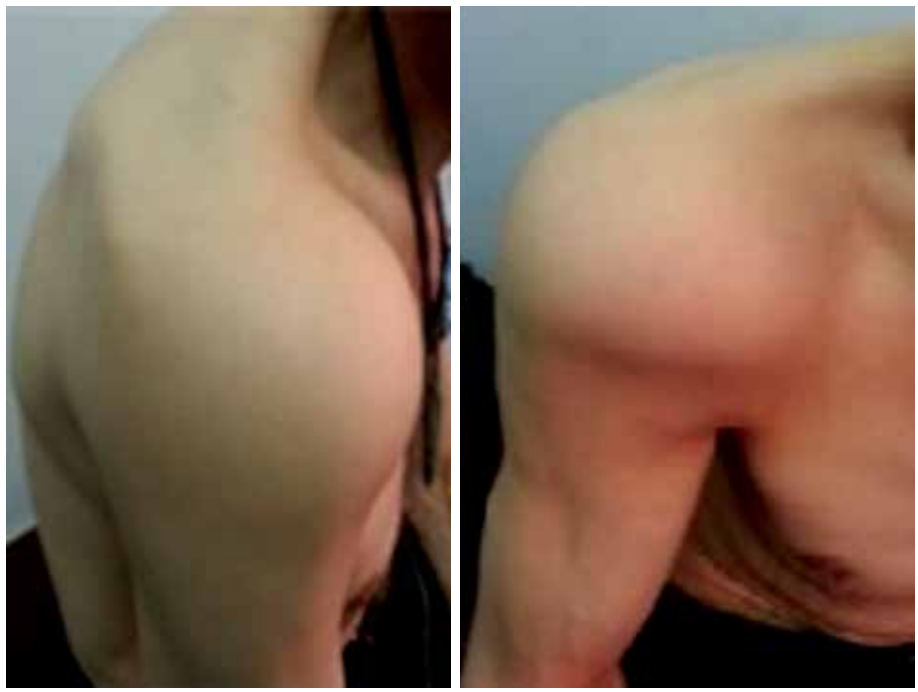

Figure 1. Swelling around right shoulder, representing subacromial-subdeltoid bursitis.

other pathological findings. He had no previous history of trauma, fever, weight loss, or night sweats. The patient was a farmer who lived in rural area and was engaged in husbandry. He defined that occasionally he had consumed raw dairy products, particularly fresh cheese. Laboratory findings included: white blood cell count 8,300/mm^3 (normal range: 3,500-11,000/mm^3), hemoglobin 15 g/dL (normal range: 13.2-17.3 g/dL), platelet count 120×10^9/L (normal range: 150-450×10^9/L), C-reactive protein (CRP) 12.3 mg/L (normal range: 0.1-5 mg/L), and erythrocyte sedimentation rate (ESR) 25 mm/hour (normal range: 0-15 mm/hour). Serum standard tube agglutination (STA) titer was of 1/640 (normal range: <1/160). Needle aspiration of the bursa yielded 10 mL of a seropurulent fluid. Blood and bursal aspiration cultures were incubated for 21 days in BACTEC 9050 (Becton Dickinson, Sparks, MD, USA). There was no bacterial growth in

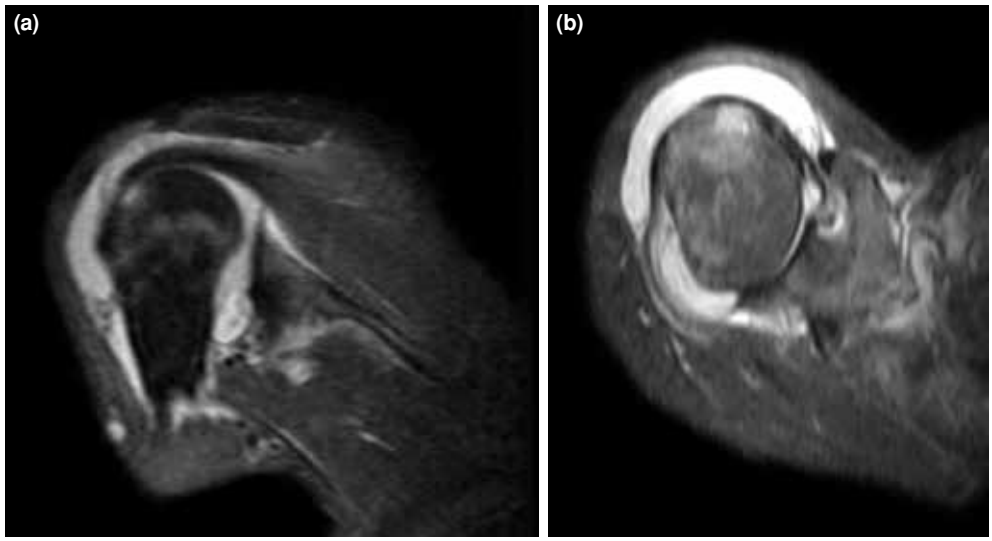

Figure 2. T2-weighted **(a)** sagittal and **(b)** coronal magnetic resonance images of right shoulder, showing subacromial-subdeltoid bursitis.

blood cultures but Brucella melitensis was isolated in bursal aspiration fluid cultures on the fourth day of the incubation. Magnetic resonance imaging of the shoulder joint demonstrated joint effusion, increased bursal fluid in subacromial-subdeltoid bursa and edema in surrounding tissues (Figure 2). Patient was treated initially with doxycycline (100 mg 2×1 per oral) and rifampicin (600 mg 1x1 per oral) for six weeks. Upon follow-up, the patient showed significant decrease in the shoulder swelling. Patient has not shown any evidence of relapse within two years of follow-up. A written informed consent was obtained from the patient.

DISCUSSION

Brucellosis is a systemic disease that can involve any organ in the human body.[2] Osteoarticular system is a common site of involvement in brucellosis. It is usually seen as sacroiliitis, spondylitis, osteomyelitis or peripheral arthritis.[7,8] Bursitis is one of the rarest forms of osteoarticular brucellosis. The incidence rate of bursitis in brucellosis patients was reported between 0.4% and 5.7%, which represents about 1.2 to 9.0% of patients with osteoarticular brucellosis.[8-10]

Brucellar bursitis was first described in 1904 by Kennedy.[11] Brucellosis has been described involving a

TABLE I

Previously published cases of brucellar bursitis in the English-language literature

Reference	Year	Patients in series (n)	Bursitis case (n)	Location of bursal involvement	Sub-species
Kennedy[11]	1904	1	1	Subdeltoid-Subacromial	Not reported
Johnson and Weed[12]	1954	4	4	Prepatellar	Brucella abortus
Rotes-Querol[17]	1957	174	3	Olecranon (2) Trochanteric (1)	Not reported
Schirger et al.[18]	1960	224	3	Prepatellar	Brucella abortus (2) Brucella suis (1)
Kelly et al.[19]	1960	36	8	Prepatellar	Brucella abortus (6) Brucella suis (2)
Ariza et al.[20]	1985	331	3	Not reported	Not reported
Mousa et al.[9]	1987	452	1	Olecranon	Not reported
Colmenero et al.[4]	1991	263	2	Olecranon	Not reported
González-Alvaro et al.[21]	1994	2	1	Olecranon (bilateral)	Brucella melitensis
McDermott et al.[22]	1994	1	1	Suprapatellar	Brucella abortus
Solera et al.[23]	1996	64	2	Prepatellar	Not reported
Davis and Broughton[24]	1996	1	1	Prepatellar	Brucella abortus
González-Gay et al.[25]	1997	1	1	Prepatellar	Brucella abortus
García-Porrúa et al.[26]	1999	75	3	Not reported	Brucella abortus
González-Gay et al.[27]	1999	158	3	Prepatellar (2) Olecranon (1)	Brucella abortus
Taşova et al.[8]	1999	238	5	Prepatellar (3) Subacromial (2)	Not reported
Guiral et al.[28]	1999	1	1	Iliopsoas	Brucella melitensis
Pourbagher et al.[10]	2006	251	13	Suprapatellar (6) Olecranon (3) Subacromial (3) Lateral malleolus (1)	Not reported
Traboulsi et al.[16]	2007	1	1	Prepatellar	Brucella melitensis
Turan et al.[29]	2009	1	1	Olecranon	Not reported
Wallach et al.[30]	2010	1	1	Prepatellar	Brucella abortus
Almajid [31]	2017	1	1	Prepatellar	Not reported

number of bursaes such as olecranon, prepatellar and subacromial. Sixty cases of brucellar bursitis have been reported in the English literature. The most common form is prepatellar bursitis and accounted for 42% (n=27) of all reported cases (Table I). Local inoculation with Brucella, which occurred during recurrent trauma from kneeling, causes most of prepatellar bursitis.[12] Subacromial-subdeltoid bursitis is one of the rarest forms of brucellar bursitis with only six cases reported in the English literature.[14] Taşova et al.[8] and Pourbagher et al.[10] reported subacromial-subdeltoid bursitis in two and three cases, respectively.

Diagnosis of brucellosis is often a challenge and depends on a large extent on clinical suspicion. Particularly in endemic areas, a clinician should consider brucellosis in the presence of non-specific signs such as fever, fatigue, sweating, hepatomegaly, and splenomegaly.[1,6] In osteoarticular cases, lower back pain and sacroiliac joint pain are the main symptoms.[7] A definite diagnosis of brucellosis is established by isolating Brucella species (spp.) from blood, bone marrow or samples of other tissues. However, positive blood culture ratio was reported only as 12 to 21%[7,8] in osteoarticular brucellosis patients. In the presented case, there was no bacterial growth in blood cultures but Brucella melitensis was isolated in bursal aspiration fluid culture. Although isolating of Brucella spp. in blood or other tissues is the gold standard in diagnosis, low growth ratio of cultures is one of the factors that makes the diagnosis of brucellosis a challenge. We recommend incubation of the bursal aspiration fluid in addition to the blood culture in the cases where brucellar bursitis is suspected. As in the current case, the routine blood markers which are used to monitor infections, such as ESR or CRP, are often redundant.[13] Serology is the most helpful laboratory test in the diagnosis and almost always positive at high titers in patients with osteoarticular involvement.[4] Taşova et al.[8] stated that 97.7% of patients with osteoarticular involvement had significantly higher Brucella antibody titers. Particularly in endemic regions, >1/160 titers of STA should be considered as brucellosis.

With or without surgical bursectomy, antibiotics are the main choice in the treatment of brucellar bursitis. Improvements in antimicrobial treatment of brucellosis have also reduced the need for surgical intervention. As in this case, a combination of doxycycline and rifampicin for 45 days was stated as an effective regimen for brucellosis treatment.[15] In this patient, a single aspiration of bursal fluid was sufficient. However, repeated aspirations may be required in the treatment of brucellar bursitis[16] in other cases.

In conclusion, a high degree of suspicion in the right clinical setting is required for the diagnosis of brucellosis. Although brucellar bursitis is a very rare form of osteoarticular involvement, the majority of patients with this complaint are evaluated in the orthopedics department in the first place. Therefore, the awareness of orthopedic surgeons about brucellosis should be increased particularly in endemic regions.

Declaration of conflicting interests

The author declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The author received no financial support for the research and/or authorship of this article.

REFERENCES

1. Young EJ. Brucella species. In: Mandell GL, Bennett GE, Dolin R, editors. Mandell, Douglas and Bennett's principles and practice of infectious diseases. 6th ed. Philadelphia: Elsevier; 2005. p. 2669-74.
2. Young EJ. An overview of human brucellosis. Clin Infect Dis 1995;21:283-9.
3. Tekkök IH, Berker M, Ozcan OE, Ozgen T, Akalin E. Brucellosis of the spine. Neurosurgery 1993;33:838-44.
4. Colmenero JD, Reguera JM, Fernández-Nebro A, Cabrera-Franquelo F. Osteoarticular complications of brucellosis. Ann Rheum Dis 1991;50:23-6.
5. Tsolia M, Drakonaki S, Messaritaki A, Farmakakis T, Kostaki M, Tsapra H, et al. Clinical features, complications and treatment outcome of childhood brucellosis in central Greece. J Infect 2002;44:257-62.
6. Hall WH. Brucellosis. In: Evants AS, Brachman PS, editors. Human bacterial infections. 2nd ed. New York: Plenum Medical Book Co.; 1991. p. 133-49.
7. Turan H, Serefhanoglu K, Karadeli E, Togan T, Arslan H. Osteoarticular involvement among 202 brucellosis cases identified in Central Anatolia region of Turkey. Intern Med 2011;50:421-8.
8. Taşova Y, Saltoğlu N, Sahin G, Aksu HS. Osteoarthricular involvement of brucellosis in Turkey. Clin Rheumatol 1999;18:214-9.
9. Mousa AR, Muhtaseb SA, Almudallal DS, Khodeir SM, Marafie AA. Osteoarticular complications of brucellosis: a study of 169 cases. Rev Infect Dis 1987;9:531-43.
10. Pourbagher A, Pourbagher MA, Savas L, Turunc T, Demiroglu YZ, Erol I, et al. Epidemiologic, clinical, and imaging findings in brucellosis patients with osteoarticular involvement. AJR Am J Roentgenol 2006;187:873-80.
11. Kennedy JC. Notes on a case of chronic synovitis, or bursitis, due to organism of the Mediterranean fever. J R Army Med Corps 1904;2:178-80.
12. Johnson EW Jr, Weed LA. Brucellar bursitis. J Bone Joint Surg [Am] 1954;36:133-9.
13. Sankaran-Kutty M, Marwah S, Kutty MK. The skeletal manifestations of brucellosis. Int Orthop 1991;15:17-9.

14. Atik OŞ. Which articles do we prefer to publish? Eklem Hastalik Cerrahisi 2018;29:1.
15. Solís García del Pozo J, Solera J. Systematic review and meta-analysis of randomized clinical trials in the treatment of human brucellosis. PLoS One 2012;7:e32090.
16. Traboulsi R, Uthman I, Kanj SS. Prepatellar Brucella melitensis bursitis: case report and literature review. Clin Rheumatol 2007;26:1941-2.
17. Rotes-Querol J. Osteo-articular sites of brucellosis. Ann Rheum Dis 1957;16:63-8.
18. Schirger A, Nichols DR, Martin WJ, Wellman WE, Weed LA. Brucellosis: experiences with 224 patients. Ann Intern Med 1960;52:827-37.
19. Kelly PJ, Martin WJ, Schirger A, Weed LA. Brucellosis of the bones and joints. Experience with thirty-six patients. JAMA 1960;174:347-53.
20. Ariza J, Gudiol F, Valverde J, Pallarés R, Fernández-Viladrich P, Rufí G, et al. Brucellar spondylitis: a detailed analysis based on current findings. Rev Infect Dis 1985;7:656-64.
21. González-Alvaro I, Estévez M, Carmona-Ortels L, Alvaro-Gracia JM, López-Bote JP, Humbria A. Osteoarticular brucellosis resembling microcrystalline arthropathy. J Rheumatol 1994;21:1783-5.
22. McDermott M, O'Connell B, Mulvihill TE, Sweeney EC. Chronic Brucella infection of the supra-patellar bursa with sinus formation. J Clin Pathol 1994;47:764-6.
23. Solera J, Espinosa A, Geijo P, Martínez-Alfaro E, Sáez L, Sepúlveda MA, et al. Treatment of human brucellosis with netilmicin and doxycycline. Clin Infect Dis 1996;22:441-5.
24. Davis JM, Broughton SJ. Prepatellar bursitis caused by Brucella abortus. Med J Aust 1996;165:460.
25. González-Gay MA, Blanco R, García-Pais MJ, Alonso P, Rodríguez A, Coira A, et al. Polymicrobial prepatellar septic bursitis caused by Brucella abortus and streptococcus agalactiae. J Clin Rheumatol 1997;3:120.
26. García-Porrúa C, González-Gay MA, Ibañez D, García-País MJ. The clinical spectrum of severe septic bursitis in northwestern Spain: a 10 year study. J Rheumatol 1999;26:663-7.
27. González-Gay MA, García-Porrúa C, Ibañez D, García-País MJ. Osteoarticular complications of brucellosis in an Atlantic area of Spain. J Rheumatol 1999;26:141-5.
28. Guiral J, Reverte D, Carrero P. Iliopsoas bursitis due to Brucella melitensis infection--a case report. Acta Orthop Scand 1999;70:523-4.
29. Turan H, Serefhanoglu K, Karadeli E, Timurkaynak F, Arslan H. A case of brucellosis with abscess of the iliacus muscle, olecranon bursitis, and sacroiliitis. Int J Infect Dis 2009;13:e485-7.
30. Wallach JC, Delpino MV, Scian R, Deodato B, Fossati CA, Baldi PC. Prepatellar bursitis due to Brucella abortus: case report and analysis of the local immune response. J Med Microbiol 2010;59:1514-8.
31. Almajid FM. A Rare Form of Brucella Bursitis with Negative Serology: A Case Report and Literature Review. Case Rep Infect Dis 2017;2017:9802532.