Fırat Tıp Dergisi 2010;15(1): 44-47

Case Report

www.firattipdergisi.com

Spondilodiscitis Due to Ankylosing Spondylitis in a Female Patient with Chronic Relapsing Brucellosis: A Case Report

Zuhal ÖZERİ, Barış NACIR, Burcu Duyur ÇAKIT[a], Meryem SARAÇOĞLU, Hatice Rana ERDEM

S.B. Ankara Eğitim ve Araştırma Hastanesi, 2. Fizik Tedavi ve Rehabilitasyon Kliniği, ANKARA, Türkiye

ABSTRACT

Ankylosing Spondylitis (AS) and brucellosis are two distinct diseases which may be involved both sacroiliac joints and intervertebral discs. Spinal involvement in both diseases may be similar sometimes, and differential diagnosis may be difficult. We reported here a female patient with concomitant AS and brucellosis who was misdiagnosed as spondylodiscitis and sacroiliitis due to brucellosis, because of similar spinal involvement.

Key words: *Ankylosing spondylitis, brucellosis, spondylodiscitis*

ÖZET

Kronik Tekrarlayan Bruselloz'lu Kadın Hastada Ankilozan Spondilite Bağlı Spondilodiskit: Vaka Sunumu

Ankilozan spondilit (AS) ve bruselloz hem sakroiliak eklemleri hem de intervertebral diskleri etkileyebilen 2 ayrı hastalıktır. Her iki hastalığıun spinal tutulumu bazen benzer ve ayırıcı tanısı zor olabilir. Biz burada benzer spinal tutulum nedeniyle yanlışlıkla bruselloza bağlı spondilit ve sakroileit tanısı konmuş AS'le birliktelik gösteren bir kadın bruselloz hastası sunduk.

Anahtar Sözcükler: *Ankilozan spondilit, bruselloz, spondilodiskit*

Brucellosis is a common and serious infectious disease in many parts of the world. The most common form of chronic and relapsing brucellosis is caused by brucella melitensis, and this species causes most of cases of brucella arthritis. The clinical manifestations and the severity of disease vary according to the responsible agent and the host. Musculoskeletal involvement tends to occur in young patients with brucellosis. Patterns of joint involvement suggest a spondyloarthropathy with axial fibrocartilaginous joints and lower extremity diarthrodial joints predominanting. Spondylitis was not common in the lumbosacral region, generally affecting older patients with chronic infection (1-5). Partly resembling the manifestations of AS, spondylitis and sacroiliitis with or without peripheral arthritis may be seen in brucellosis (1).

Ankylosing spondylitis (AS) is a prototype of the seronegative spondyloarthropathies and characterized by inflammation of the axial skeleton with sacroiliac joint involvement as its hallmark with or without peripheral joint involvement and extraarticular features (6). Spondylodiscitis may develop at any time during the course of AS and may be asypmtomatic. We report here the case of a female patient with concomitant AS and brucellosis who was misdiagnosed as spondylitis and sacroiliitis due to brucellosis, because of similar spinal involvement.

CASE REPORT

A 30 years old female patient was admitted to our outpatient clinic because of her buttock and low back pain lasting for 5 years. In 1997 she was diagnosed and treated as Brucellosis. She had relapses in 1997, 1999 and in 2000. At that times she complained increasing pain in her low back and buttocks which became worse by movement and relieved by rest. In 1999, she had undergone magnetic resonance imaging (MRI) examination which showed sacroiliitis bilaterally and spondylitis, and she was thought to have sacroiliitis and spondylitis due to brucellosis (Figure 1).

Our physical examination revealed limitation of lumbar spinal movement, spasm of lumbar paravertebral muscles, decrease in lumbar lordosis. Extreme tenderness was on the spinous process of L_2-L_3 vertebra with palpation. Sacroiliac compression tests, Gaenslen's, Mennel's and Patrick's tests were positive bilaterally. Lumbar Schober test was diminished to 1.5 cm, wall to tragus distance was 10 cm and chest expansion was 1.5 cm. There was no swelling or tenderness of peripheral joints. There were no objective neurological signs. The patient was hospitalized.

Her white blood cell count was $7.2x10^9$ µL, with 56% neutrophils and 24% lymphocytes; hemoglobin was 11.7g/dl, hematocrit %, erythrocyte sedimentation rate (ESR) was 31 mm/h. C reactive protein (CRP) level of 9.45 mg/l (normal: 0-6 mg/dl) and rheumatoid factor was negative. Blood urea, creatinine levels and liver function tests were normal.

Blood cultures were negative, a rose Bengal test was 1/160, Wright's seroagglutination test was 1/160, Coombs test for brucella was 1/160.

[a] Corresponding Adress: Dr. Burcu Duyur ÇAKIT, S.B. Ankara Eğit. ve Araş. Hast., 2. Fizik Tedavi ve Rehabilitasyon Kliniği, ANKARA, Türkiye
Tel: +90 312 2847834 e-mail: burcudcakit@yahoo.com

Plain anteroposterior radiographs of lumbar spine showed spinal syndesmophytes and grade 3 sacroiliitis bilaterally (Figure 2). In 2002 MRI of lumbar spine showed L_2-L3 spondylodiscitis and L5-S1 posterocentral disc protrusion (Figure 3). Computerized tomography of sacroiliac joints demonstrated minimal narrowing and destruction bilaterally (Figure 4). HLA typing showed that HLA B-27 was positive. Abdominal ultrasonogram was normal.

In the light of these clinical findings, we decided that the patient had a delayed diagnosis of AS and thus we started 2000 mg/day sulfasalazin and 15 mg/day meloxicam treatment. After 3 months her back pain reduced. Her ESR was 26 mm/h and her CRP level was 2.3 mg/dl.

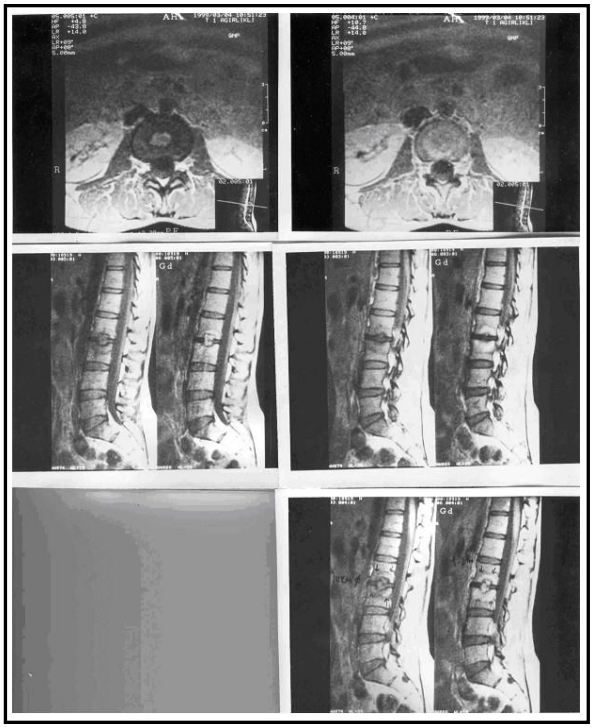


Figure 1. *Sagittal and axial magnetic resonance images of the lumbar spine showing L3-L4 intervertebral spondilodiscitis in 1999.*

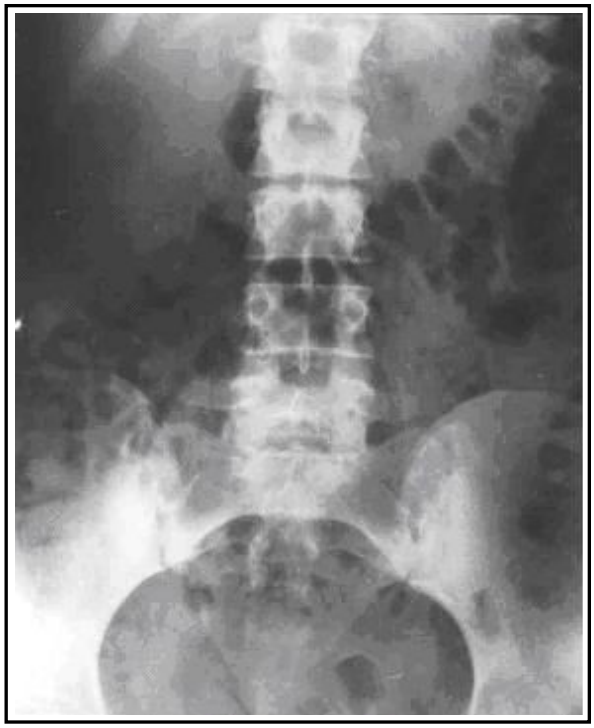
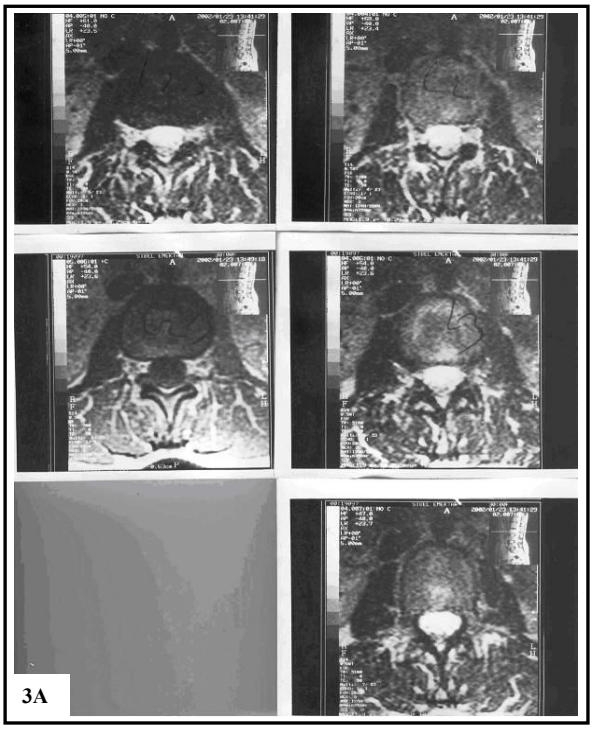


Figure 2. *Anterior-posterior radiograph of the lumbar spine showing bilateral symmetric sacroiliiitis.*



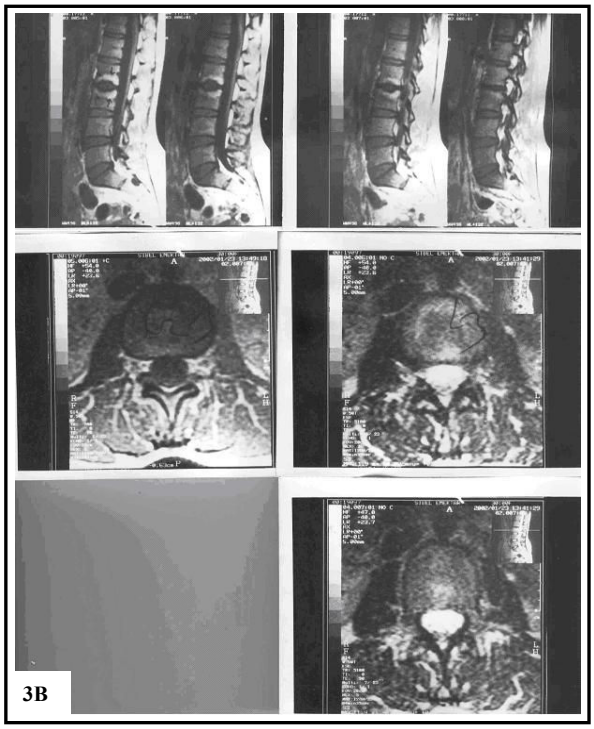


Figure 3. *Sagittal and axial magnetic resonance images of the lumbar spine showing L3-L4 intervertebral spondilodiscitis in 2002.*

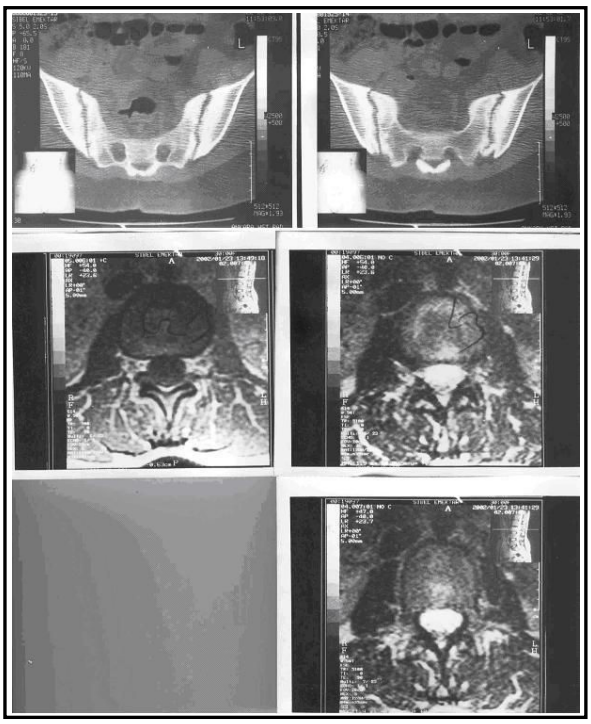


Figure 4. *Computerized Tomography images of the sacroiliac joints showing bilateral symmetric sacroiliitis.*

DISCUSSION

Brucellosis often affects the musculoskeletal system and is included in the differential diagnosis of variety of clinical pictures (7, 8). Osteoarticular complications of brucellosis occur in 10%-85% patients (9-12). The spine is the most common affected site and back pain is the most common clinical manifestation of brucellosis (11, 13-15). Colmenero et al. found that; spondylitis occured in 58%, sacroiliitis in 45% of their patients who had osteoarticular complications of brucellosis (16).

Sacroiliitis of brucellosis usually occurs unilaterally in young patients, and is asymmetric in cases with bilateral involvement. (1, 7, 12, 17). In brucellosis, there is no radiologic progression changes and ankylosis in sacroiliac joint and it is resolved the problem without residual damage (1, 7, 8). Taşova et al. reported that sacroiliitis has been determined as the most frequently osteoarticular complication of brucellosis in Turkey. They found that concomitant sacroiliitis and spondylitis were only in 6 of 238 patients (7). Sacroiliitis is significantly associated with lumbar or sacral spondylitis in the elderly patients (4).

Brucellar spondylitis may be difficult to diagnose and can be concomitant with connective tissue diseases and spondylarthropathies and changes in plain radiographs can be difficult to differentiate from degenerative diseases (8-10). The incidence of spondylitis reported in the literature ranges from 10% to 50%. İt is seen especially in elderly men over 50 years of age (7, 14, 18, 19).

If both sacroiliitis and spondylitis are seen in young patients, the noninfectious causes of sacroiliitis and spondylitis should be considered. Most patients with brucellar spondylitis and sacroiliitis respond to antibiotic treatment (10, 14). Sacroiliitis and spondilitis are not related to the presence of HLA-B27 antigen (4) but HLA B27 was positive in our case. Additionally our patient has bilateral simetric sacroiliitis and spondilitis which did not respond to antibiotic treatment.

Destructive lesions of spondylodiscitis involving an intervertebral disc space and adjoining vertebral bodies are seen in AS. Radiological estimates of the prevalence of spondylodiscitis are reported in a range of 1-28%. Destructive change at the discovertebral junction may be predominately peripheral, central or both. Spondylodiscitis in AS is often asymptomatic (20, 21). But if an AS patient presents pain increasing after movements and improving at rest, spondilodiscitis should be considered (21). The low back pain in brucellosis does not reduce with rest and exercise (8). As in our case, the altering character of the inflammatory low back pain in the presence of spondilodiscitis in the patients with spondylitis leads to difficulties in differential diagnosis.

MRI plays an important role in the diagnosis, assesment, and management of patients with spondylitis (10). MRI is noninvasive, involves no radiation exposure and provides excellent multiplanar views of osseous and soft tissues (2, 22). In the spine, MRI findings of AS includes sacroiliitis, spondylodiscitis, pseudoarthrosis, fractures, atlantoaxial subluxation, ossification of ligaments and cauda equina syndrome. The spectrum of infections of the spinal column includes vertebral osteomyelitis, discitis and paravertebral and epidural phlegmon or abscess (2).

After successfull treatment of infectious spondylitis, the MRI findings slowly change to normal (2). But in our case, the spine involvement had the characteristics of spondylarthropathy, the radiologic progress was continued in spite of the treatment, bilateral and symmetric sacroiliitis was present and HLA-B27 was positive. In the light of these clinical findings, the lesion at L_2-L_3 vertebrae was accepted as ‘spondilodiscitis’ associated with AS.

In conclusion, in endemic regions, coexistance with AS and brucellosis should always be in mind of the physicians and diagnosis of brucellosis alone should not be regarded as satisfying all the time especially in cases resistant to trea.

REFERENCES

1. Gotuzzo E, Alarcon GS, Bocanegra TS, Carillo C, Guerra JC, Rolando I, Espinoza LR. Articular involvement in human brucellosis: a retrospective analysis of 304 cases. Semin Arthritis Rheum 1982;12:245-255.

2. Oostveen JCM, Van De Laar MAFJ. Magnetic resonance imaging in rheumatic disorders of the spine and sacroiliac joints. Semin Arthritis and Rheum 2000; 30:52-69.

3. Gotuzzo E, Seas C, Guerra JG, Carrillo C, Bocanegra TS, Calvo A, Castaneda O, Alarcon GS. Brucellar arthritis: a study of 39 Peruvian families. Ann Rheum Dis 1987;46:506-509.

4. Ariza J, Pujol M, Valverde J, Nolla JM, Rufi G, Viladrich PF, Corredoira JM, Gudiol F. Brucelar sacroiliitis: Findings in 63 episodes and current relevance. Clin Infect Dis 1993;16:761-765.

5. Ardic F, Kahraman Y, Soyupek F. Co-existence of ankylosing spondylitis and brucellosis in a female patient. The Pain Clinic 2004; 16:363-367.

6. Van Der Linden, Van Der Heijde. Spondyloarthropathies. In: Ruddy S, Harris ED, Sledge CB, ed. Kelley's Textbook of Rheumatology. Philadelphia, W.B. Saunders Company, 2001:9:69:1039-1053.

7. Taşova Y, Saltoğlu N, Şahin G, Aksu HSZ. Osteoarticular involvement of brucellosis in Turkey. Clin Rheumatol 1999;18:214-219.

8. Ozgocmen S, Ardicoglu A, Karakoc E, Kiris A, Ardicoglu O. Paravertebral abscess formation due to brucellosis in a patient with ankylosing spondylitis. Joint Bone Spine 2001;68:521-524.

9. Duyur B, Erdem HR, Ozgocmen S. Paravertebral abscess formation and knee arthritis due to Brucellosis in a patient with rheumatoid arthritis (Case report). Spinal Cord 2001;39:554-556.

10. Solera J, Lozano E, Martinez-Alfaro E, Espinosa A, Castillejos ML, Abad L. Brucelar Spondylitis: Review of 35 cases and literature survey. Clin Infect Dis;29:1440-1449.

11. Sankaran-Kutty M, Marwah S, Kanan-Kutty M. The skeletal manifestations of brucellosis. Int Orth 1991;15:17-19.

12. Al-Eissa YA, Kambal AM, Alrabeeah AA, Abdullah AMA, Al-Jurayyan NA, Al-Jishi NM. Osteoarticular brucellosis in children. Ann Rheum Dis 1990;49:896-900.

13. Tekkok IH, Berker M, Ozcan OE, Ozgen T, Akalın E. Brucellosis of the spine. Neurosurgery 1993;33:838-844.

14. Rajapakse CNA, Al-Aska KA, Al-Orainey I, Halim K, Arabi K. Spinal brucellosis. Br J Rheum 1987;26:28-31.

15. Mohan V, Gupta RP, Marklund T, Sabri T. Spinal brucellosis. Int Orth 1990;14:63-66.

16. Colmenero JD, Reguera JM, Fernandez-Nebro A, Cabrera-Franwuela F. Osteoarticular complications of brucellosis. Ann Rheum Dis 1991;50:23-26.

17. Alarcon GS, Bocanegra TS, Gotuzzo E, Espinoza LR. The arthritis of brucellosis: A Perspective one hundred years after Bruce's discovery (Editorial). J Rheum 1987;14:1083-1085.

18. Ariza J, Gudiol F, Valverde J, Pallares R, Fernandez-Viladrich P, Ruffi G, Espadaler L, Fernandez-Nogues F. Brucellar spondylitis: A detailed analysis based on current findings. Rev Infect Dis 1985;7:656-664.

19. Colmenero JD, Cisneros JM, Orjuela DL, Pachon J, Garcia-Portales R, Rodriguez-Sampedro F, Juarez C. Clinical course and prognosis of brucella spondylitis. Infection 1992;20:38-42.

20. Kabasakal Y, Garrett SL, Calin A. The epidemiology of spondylodiscitis in ankylosing spondylitis-A controlled study. Br J Rheum 1996;35:660-663.

21. Rasker JJ, Prevo RL, Lanting PJH. Spondylodiscitis in ankylosing spondylitis, inflammation or trauma? Scand J Rheumatol 1996;25:52-57.

22. Battafarano DF, West SG, Rak KM, Fortenbery EJ, Chantelois AE. Comparison of bone scan, computed tomography, and magnetic resonance imaging in the diagnosis of active sacroiliitis. Semin Arthritis Rheum 1993;23:161-176.

Kabul Tarihi: 18.10.2009