

**CASE REPORT**

## **Brucella spondylodiscitis: Multifocal involvement in thoracic and lumbar areas; a rare case**

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### **ABSTRACT**

Brucellosis is a zoonosis that can affect many organs or systems. Musculoskeletal system is the most commonly affected site in brucellosis and the frequency of musculoskeletal system involvement varies between 2% and 53%. Multifocal spinal involvement is rare. Here, it was aimed to present our case with brucellar spondylodiscitis involving the thoracic and lumbar regions simultaneously. MRI is a useful imaging method in the diagnosis of multifocal spinal infection caused by brucellosis. Especially in endemic regions, at least two serological tests should be used in the diagnosis of brucellosis. Brucellosis should be absolutely kept in mind for the differential diagnosis of the elderly patients with complaint of longstanding thoracic and back pain in the regions where brucellosis is endemic. *J Microbiol Infect Dis* 2015;5(3): 129-132

**Key words:** Brucellosis, spinal infection, spondylodiscitis.

## **Brucella spondilodiskiti: Torasik ve lomber bölgenin multifokal tutulumu; nadir bir olgu**

### **ÖZET**

Bruselozis bir çok organ ve dokuları tutabilen bir zoonotik enfeksiyondur. İskelet sistemi bruselloziste en sık etkilenen bölge olup, sıklığı % 2 ile % 53 arasında değişmektedir. Multifokal spinal tutulum nadirdir. Bu yazıda torakal ve lomber bölgeyi aynı anda tutan brucella spondilodiskit olgusunun sunulması amaçlanmıştır. Brusellaya bağlı multifokal spinal enfeksiyonun tanısı MRI ile konulabilir. Özellikle endemik bölgelerde brusellozis tanısında en az iki adet serolojik test yöntemi kullanılması gerekmektedir. Endemik bölgelerde uzun zaman devam eden sırt ve bel ağrısı şikayeti olan hastalarda ayırıcı tanıda mutlaka brusellozis düşünülmelidir.

**Anahtar kelimeler:** Brusellozis, spinal enfeksiyon, spondilodiskitis.

### **INTRODUCTION**

Brucellosis is a systemic infection caused by a bacterium of the brucella genus which can involve various systems.<sup>1,2</sup> Osteoarticular involvement is one of the most commonly seen complications of brucellosis and it affects 10-85% of the patients. Osteoarticular involvement in brucellosis is most commonly encountered as followings: spondylitis, sacroiliitis, osteomyelitis, peripheral arthritis, bursitis, and tenosynovitis.<sup>2</sup>

Spinal infection due to brucellosis causes morbidity resulting from infectious disease in the infested regions of the world and particularly in the

rural areas including our country.<sup>3</sup> Spondylodiscitis is most commonly seen in the lumbar, cervical and thoracic regions, respectively.<sup>4</sup> Multifocal involvement is very rare in brucellar spondylodiscitis. Here, it was aimed to present our case with brucellar spondylodiscitis involving the thoracic and lumbar spine regions simultaneously.

### **CASE**

A 75-year-old male patient presented to the neurosurgery department of our hospital with a complaint of longstanding pain in the back, lower-back regions, and right leg for 6 months. He reported tenderness

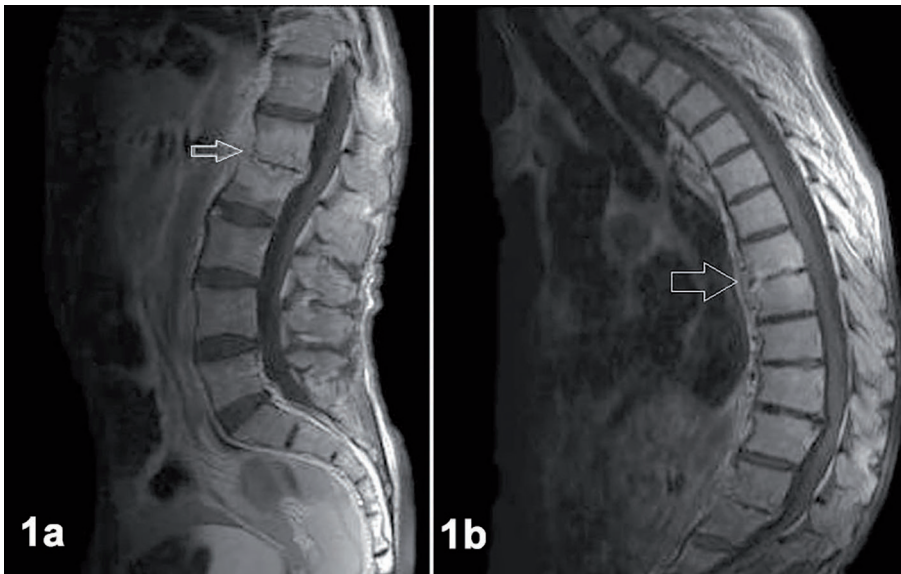
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in the back and lower back on palpation and there was severe restriction of back movements due to back pain. On physical examination kyphotic deformation was detected, there was no neurological deficit. In laboratory tests, leukocyte count, erythrocyte sedimentation rate (ESR) and C reactive protein (CRP) values were within normal ranges. On lateral radiography views, there were erosive changes in the thoracic and lumbar vertebra. Spinal Magnetic Resonance Imaging (MRI) demonstrated narrowing

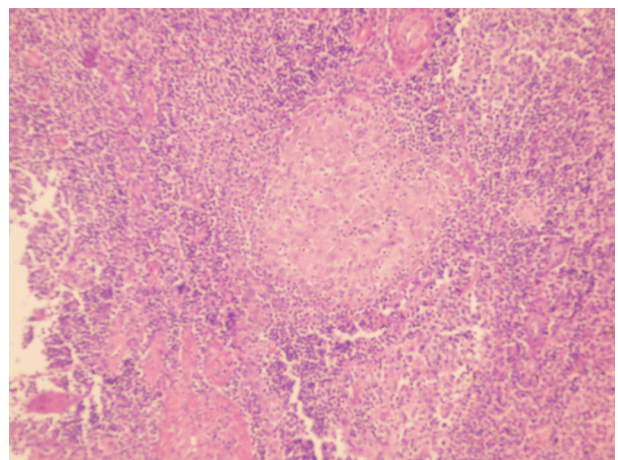
with irregular vertebral endplates affecting simultaneously the thoracic and lumbar disc spaces. MRI examination revealed contrast enhancement in intervertebral disc and vertebral corpus of Th8- Th9 and L1-L2 levels (Figure 1a, 1b). It was reported MRI findings may compatible with a metastatic infiltrative process or spondylodiscitis. Whole body scintigraphy revealed degenerative changes in Th8-Th12 and L1- L2 but no pathological activity favoring metastasis was observed.



**Figure 1a.** MR scan sagittal view of the thoracic spine: After contrast administration there is contrast enhancement on vertebral bodies and intervertebral disc at the T8-T9 levels.

**Figure 1b.** MR scan sagittal view of the lumbar spine: Similar changes to the thoracic spine suggesting spondylodiscitis were also evident at the L1-L2 levels.

Due to suspicion of tuberculosis patient have been consulted by chest specialist. A tuberculin skin test was negative at 72 h and the chest x-ray did not detect pulmonary lesions. Due to suspicion of brucellosis standard tube agglutination (STA) was done and test titre was 1/80 and the the clinician didn't need to perform any further diagnostic test for brucellosis. A total L1-2 laminectomy was performed on the patient. No complications occurred and the patient's pain was decreased, so the patient was discharged. Histopathological examination of the biopsy material revealed discitis (Figure 2). Ciprofloxacin was started for 15 days. The patient admitted to the neurosurgery department three months after the operation with a progressive back and lower back pain. MRI examination disclosed contrast enhancement in the vertebral corpus of Th8-Th9 and L1-L2, and the patient was referred to the infections department for investigation discitis etiology.



**Figure 2.** Chondroid tissue destroyed by plasma cells, lymphocytes and polymorphonuclear leukocytes. (HE x 40)

Patient referred to our infectious diseases outpatient clinic with a history of fatigue, lower back and back pain in the previous ten months which had decreased after the surgical operation and re-appeared in the previous month. He also reported continue fever, pain and weight loss initially. In his history, habitation in a rural area and stockbreeding were noted. At the physical examination, the patient was afebrile and his systemic examination was normal. Since the patient lived in the rural area and he reported consumption of dairy products, he was suspected to have brucellosis. STA test result and Coombs' brucella agglutination tests were 1/80 and 1/320, respectively. The patient received a combination of rifampicin (600 mg/day, p.o.) and doxycycline (200 mg/day, po) with a diagnosis of multifocal brucellar spondylodiscitis. *Brucella* spp. couldn't be detected in blood cultures. The patient was negative for brucella IgM antibodies and positive for IgG antibodies. Antibiotics were continued for a total of three months. The back and lower back pain reduced progressively with medical treatment.

At 3-months follow-up, the patient was admitted to the infections department with recurrence of pain, fatigue. In laboratory tests: leukocyte count, 16.600/ $\mu$ l (4-11 x 10<sup>3</sup>/ $\mu$ l); CRP 66.8 (0-8 mg/L); and serum antibody titre was 1/320 in Coombs' brucella agglutination test. *Brucella* couldn't be detected in blood cultures. MRI examination revealed contrasted areas in the vertebral corpus of Th8-Th9 and L1-L2. The patient resumed the combination antibiotic treatment. His symptoms improved and the treatment was continued for a total of 3-months. At the end of 3-month follow-up, radiological and clinical recovery was obtained.

## DISCUSSION

Brucellar spondylodiscitis constitutes 6-58% of brucellosis osteoarticular involvements. In the spinal system, the lumbar area (60%), thoracic area (19%), and cervical area (12%) are mostly affected.<sup>4-6</sup> In the literature, multi-focal involvement in the spinal area was observed to occur in 3-14% of all cases.<sup>4-6</sup> To the best of our knowledge, 14 similar cases with non-contiguous multi-focal involvement were previously reported in the literature (Table 1).<sup>5,7-9, 11-16</sup>

Differential diagnosis between multi-focal brucellar spondylodiscitis and both degenerative disc diseases and tuberculosis is a major diagnostic problem. In case of presence of vertebral destruction, collapse or multifocal vertebral involvement, it is important to make differential diagnosis between tuberculous spondylodiscitis and brucellar spon-

ylodiscitis since it is possible to prevent complications in these diseases with an appropriate initial treatment.<sup>7</sup> In multi-focal brucellar spondylodiscitis, increased uptake of the involved area could be detected on bone scintigraphy. MRI also allows this diagnosis as it can image the whole spine. For diagnosis, MRI is more sensitive than scintigraphy and provides advantages for detecting disco-vertebral, soft tissue, and epidural involvement. MRI is also efficient in detecting multi-focal involvement, as well as, in diagnosing or excluding potential complications.<sup>6,8</sup>

In brucellar spondylodiscitis, the abnormal results of hematological and biochemical tests are mostly mild and non-specific.<sup>8</sup> The diagnosis of brucellosis is frequently made by serological methods and in most of the cases STA titre is 1  $\geq$  1/160.10 Colmenero et al. reported STA titres <1/160 in 32.3% of osteomyelitis cases caused by brucella. Therefore, they recommended at least two serological tests to increase the sensitivity of the serologic evaluation in focal complications of brucellosis.<sup>6</sup>

Cases with lower STA titres or who are serologically negative for brucella might be detected with blood culture. Pourbagher et al. reported that blood culture was positive in 38.5% of the patients with brucellar spondylodiscitis.<sup>7</sup> Similarly, Yilmaz et al. isolated brucella species from blood cultures of 32% of their patients followed-up due to brucellar spondylodiscitis.<sup>17</sup> Bozgeyik et al. reported that the diagnosis of brucellar spondylodiscitis was made based on clinical findings, serological tests and detection of the brucella in blood cultures if it was growth. They also reported that blood culture positivity rate 41% of their patients with brucellar spondylodiscitis.<sup>18</sup> We couldn't isolated brucella species from blood culture and our patient was diagnosed with clinical, serological and radiological (MRI) findings.

Insufficient microbiological techniques and the therapies previously used can be resulted with failure to detect the bacteria in blood cultures. It becomes difficult to isolate the bacterial pathogen as the disease progresses.<sup>7</sup> Therefore; cause of failure to growth organism by blood culture may result from our patients' disease duration for ten months and using antibiotics after the operation. Similarly Mrabet et al and Raptopoulou et al reported failure to growth organism by blood culture in their multi-focal involved brucellar spondylodiscitis patients.<sup>14,5</sup>

Optimal treatment decisions, including antibiotic regimen, the duration of antibiotic therapy, and

surgical procedure type, remain debatable. The recommended treatment plan includes doxycycline (200 mg/day) with rifampicin (15-20 mg/day) or doxycycline (200 mg/day) with streptomycin (1g/day). The optimal treatment duration has not been clearly established, but at least three to 6-months of treatment is recommended.<sup>2,4</sup> Nevertheless, there is not any standard therapy for osteoarticular brucellosis.<sup>1,19</sup> In the early phases of the disease, medical treatment could be efficient but in older patients (50-60 years old) it might not provide satisfactory results. A surgical procedure might be necessary for cases with neurological deficits and patients with long-term complaints. Surgery may provide fast recovery from symptoms and prevent the worsening of clinical and neurological complications.<sup>6,20,21</sup>

Consequently, here, we aimed to present our case with brucellar spondylodiscitis involving the thoracic and lumbar spine regions simultaneously. In endemic regions like our country, brucellosis should be absolutely kept in mind for the differential diagnosis of the elderly patients presenting with complaint of longstanding thoracic and back pain. MRI is an important method in the differential diagnosis and follow-up of multifocal spinal involvement in brucellosis. In multifocal vertebral involvements, it is important to consider brucellosis for the differential diagnosis.

## REFERENCES

- Young EJ. Brucella species. In: Mandell GL, Bennett JE, Dolin R, eds. Principles and practice of infectious diseases. Philadelphia, PA: Churchill Livingstone, 2000:2386-2393.
- Arkun R, Mete BD. Musculoskeletal brucellosis. *Semin Musculoskelet Radiol* 2011;15:470-479.
- Turgut M, Turgut AT, Koşar U. Spinal brucellosis: Turkish experience based on 452 cases published during the last century. *Acta Neurochir (Wien)* 2006;148:1033-1044.
- Chelli Bouaziz M, Ladeb MF, Chakroun M, Chaabane S. Spinal brucellosis: a review. *Skeletal Radiol* 2008;37:785-790.
- Raptopoulou A, Karantanas AH, Pomboulidis K, et al. Brucellar spondylodiscitis: noncontiguous multifocal involvement of the cervical, thoracic and lumbar spine. *Clin Imaging* 2006;30:214-217.
- Colmenero JD, Ruiz-Mesa JD, Plata A, et al. Clinical findings, therapeutic approach, and outcome of brucellar vertebral osteomyelitis. *Clin Infect Dis* 2008;46:426-433.
- Pourbagher A, Pourbagher MA, Savas L, et al. Epidemiologic, clinical, and imaging findings in brucellosis patients with osteoarticular involvement. *AJR Am J Roentgenol* 2006;187:873-880.
- Chelli Bouaziz M, Bougamra I, Kaffel D, et al. Non contiguous multifocal spondylitis: an exceptional presentation of spinal brucellosis. *Tunis Med* 2010;88:280-284.
- Samra Y, Hertz M, Shaked Y, et al. Brucellosis of the spine. A report of 3 cases. *J Bone Joint Surg Br* 1982;64:429-431.
- Turunç T, Demiroğlu YZ, Alişkan H, et al. Brucellosis in cases of end-stage renal disease. *Nephrol Dial Transplant* 2008;23:2344-2399.
- Baqer MM, Qurtom MA, Abdulhadi Ali Al-Ajmi J, et al. Multifocal brucellosis spondylodiscitis. *Clin Nucl Med* 2002;27:842-843.
- Shalchian S, de Wispelaere F. Brucella spondylodiscitis with multiple-level involvement, an unusual clinical presentation. *Eur J Neurol* 2007;14:1-2.
- Solera J, Lazano E, Martinez-Alfaro E, et al. Brucellar spondylitis: review of 35 cases and literature survey. *Clin Infect Dis* 1999;29:1440-1449.
- Mrabet D, Mizouni H, Khiari H, et al. Brucellar spondylodiscitis affecting non-contiguous spine levels. *BMJ Case Rep* 2011:1-4.
- Charalambides C, Papademetriou K, Sgouros S, Sakas D. Brucellosis of the spine affecting multiple non-contiguous levels. *Br J Neurosurg* 2010;24:589-591.
- Zorpala A, Skopelitis E, Thanos L, et al. An unusual case of brucellar spondylitis involving both the cervical and lumbar spine. *J Clin Imaging* 2000;24:273-275.
- Yılmaz E, Parlak M, Akalın H, et al. Brucellar spondylitis review of 25 cases. *J Clin Rheumatol* 2004;10:300-307.
- Bozgeyik Z, Ozdemir H, Demirdag K, et al. Clinical and MRI findings of brucellar spondylodiscitis. *Eur J Radiol* 2008;67:153-158.
- Buzgan T, Karahocagil MK, Irmak H, et al. Clinical manifestations and complications in 1028 cases of brucellosis: a retrospective evaluation and review of the literature. *J Infect Dis* 2010;14:469-478.
- Alp E, Doganay M. Current therapeutic strategy in spinal brucellosis. *Int J Infect Dis* 2008;12:573-577.
- Ekici MA, Ozbek Z, Gökoğlu A, Menkü A. Surgical management of cervical spinal epidural abscess caused by Brucella melitensis: report of two cases and review of the literature. *J Korean Neurosurg Soc* 2012;51:383-387.