# Rare disease

# Brucellar spondylodiscitis affecting non-contiguous spine levels

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## Summary

Brucellosis is a zoonosis that affects several organs. The spine is the most common site of musculoskeletal involvement. However, multiple-level spinal involvement is rare in brucella spondylodiscitis. The authors report a case of a 56-year-old male shepherd who had developed a spondylodiscitis affecting simultaneously the cervical, thoracic and lumbar regions. The diagnosis was established by using MRI after the brucella-agglutination test was found to be positive. A high degree of suspicion in the diagnosis of brucellar spondylodiscitis is essential to reduce the delay for the treatment. Thus, it should be essentially included in the differential diagnosis of longstanding cervical, thoracic or back pain, particularly in regions where brucellosis is endemic. Screening serological tests for brucella should be used more widely in cases with low index of suspicion, especially in endemic areas.

#### **BACKGROUND**

Brucellosis is a zoonosis of worldwide distribution and remains an important public health problem in some countries, especially in the Mediterranean region. Vertebral involvement in brucellosis is a complication of variable occurrence in the literature. Non-contiguous synchronous multifocal spinal involvement is very exceptional. We describe here a patient who has developed brucellar spondylodiscitis affecting simultaneously the cervical, thoracic and lumbar regions, and has responded well to medical treatment and conservative management.

## **CASE PRESENTATION**

A 56-year-old male shepherd was admitted to our hospital with a 1-year history of cervical and low back pain radiating to both of his legs from the buttock to heel with diffuse abdominal pain and arthromyalgia. The patient mentioned fever at 38°C before lumbar pain started. He has also reported night sweats and a weight loss of approximately 4 kg during the last 6 months. Sneezing typically aggravated the pain. On physical examination, anterior lumbar flexion was limited with pain in sitting and walking position. Lasegue test was positive at 40° and 50° on the left and right sides, respectively. The patient had also painful percussion of the thoraco-lumbar junction spine and passive mobilisation of the neck with decreased range of motion. Paravertebral muscles tenderness and contraction were found. No neurological abnormality was noted except hyperesthesia in the right S-1 dermatome. There was no splenomegaly or hepatomegaly and no masses were palpable in the abdomen.

## **INVESTIGATIONS**

Routine laboratory tests revealed white cell count at 3200/mm<sup>3</sup> with 46% neutrophils and lymphocytes at

1320/mm³; haemoglobin was 14 g/dl. Elevated erythrocyte sedimentation rate (ESR) at 90 mm at the first hour and a C reactive protein (CRP) at 112 mg/l were noted. Blood cultures grew no organism. The rest of the blood biochemistry profile was normal. Tests for hepatitis B and C viruses were negative.

Plain radiographs of the spine revealed discovertebral involvement at the C6-C7, Th10-Th11 and L1-L2 intervertebral spaces. On lateral views, we disclosed mild erosive changes of the lower cartilage endplate of Th10 and L1 vertebral bodies (figure 1). Spinal MRI demonstrated narrowing with irregular vertebral endplates affecting simultaneously the cervical (C6-C7), thoracic (Th10-Th11) and lumbar disc spaces (L1-L2). Areas of hypointensity in the intervertebral disc were noted, whereas the adjacent vertebral bodies appeared as hypointense on T1-weighted images and hyperintense on T2-weighted images. Changed signal intensity and heterogeneous contrast enhancement of vertebral bodies and intervertebral disc were noted (high signal intensity of the disk on T2-weighted images and its contrast enhancement on T1-weighted images is typical for an acute inflammatory process) (figures 2 and 3). We detected paravertebral abscesses at Th9-Th10 and at L1-L2 vertebral levels (figure 4). Left psoas abscess formation was also found (figure 4).

## **DIFFERENTIAL DIAGNOSIS**

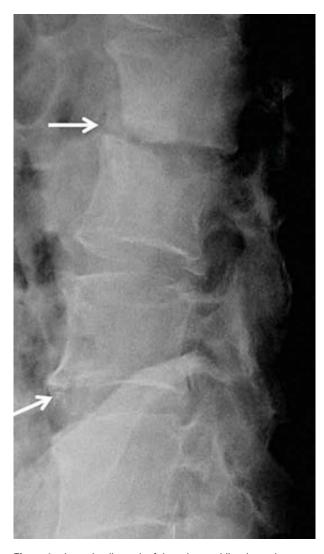
The first diagnosis was a multifocal infectious spondylodiscitis.

A tuberculin skin test was negative at 72 h and the chest x-ray did not detect pulmonary lesions. We first thought about Pott's disease because the patient lives in endemic areas of tuberculosis. Needle discovertebral biopsy of the L1-L2 level was required for definite

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**Figure 1** Lateral radiograph of the spine: multilevel anterior endplate erosive changes (arrows) suggestive of an infectious aetiology with no evidence of disc bulging.

diagnosis. Histopathology examination of a specimen revealed chronic osteitis without granulomatous inflammation or caseous necrosis.

#### **TREATMENT**

Since the patient had declared a history of unpasteurised dairy products consumption, especially white cheese and milk, the diagnosis of brucellosis was suspected. Investigations were then conducted to confirm this diagnosis. Rose Bengal test was positive and Wright agglutination test for brucella was also positive at a titre of 1/820.

Promptly, spine immobilisation with orthosis was considered justifiable for our patient. He received also a combination of antibiotics: rifampicine (900 mg/day) and doxycycline (200 mg/day) for 3 months followed by doxycycline alone for 6 months. The paravertebral and paraspinal abscesses disappeared under this antibiotic treatment and we needed a percutaneous drainage to evacuate the left psoas abscess.

## **OUTCOME AND FOLLOW-UP**

Outcome was favourable. The cervical and low-back pain reduced progressively during the course of the medical treatment. The brucella serum antibody titre was 1/320 4 weeks after the beginning of antibiotic treatment.

No relapse was seen. Almost all radiological findings disappeared at the end of 18-months' follow-up without any further treatment. Radiograph imaging showed sclerosis and reactive osteophyte formation indicating the healing phase of the inflammatory tissue at the cervical, dorsal and lumbar levels.

## **DISCUSSION**

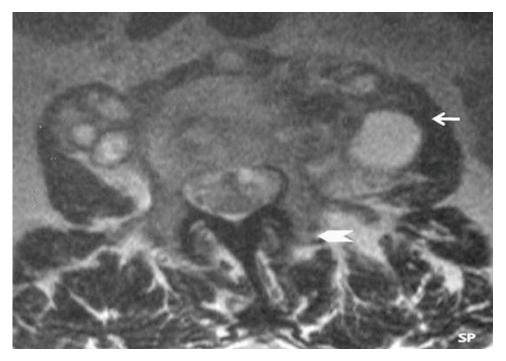
Brucellosis is an infectious disease spread by consumption of non-pasteurised milk products or through contact with infected animals. Spinal involvement is one of the most serious complications and the reported incidence varies from 2% to 60%. The lumbar area is the most frequently



Figure 2 Spondylodiscitis C6-C7. (A) T1 sagittal image. Cervical spine. Abnormal low signal in the vertebral bodies of C6 and C7. The endplates show destruction. Soft tissue masses extending both anterior and posterior to the vertebral bodies. The posterior mass is displacing the cord. (B) T1 contrast-enhanced sagittal image. Cervical spine. The marrow shows contrast enhancement, as does the disk and the mass in the anterior epidural space and the mass anterior to the spine. The diffuse enhancement of the soft tissue masses indicates that these are phlegmonous masses rather than abscesses. (C) T2 sagittal image, cervical spine. The vertebral bodies and The soft tissue phlegmon show heterogeneous high signal.



Figure 3 MR examination of the low dorsal and lumbar spine: bone oedema of the Th10, Th11, L1 and L2 endplates and paravertebral and epidural abscess in L1-L2 level and epidural thickening in Th10-Th11 level. (A) Sagittal spin-echo T1-weighted. (B) Sagittal STIR-weighted. (C) Sagittal gadolinium-enhanced T1-weighted with fat saturation.



**Figure 4** MR examination of the lumbar spine, axial spin-echoT2-weighted: epidural abscess formation associated with cord compression (arrowhead). Psoas abscesses at L1-L2 vertebral level were also found (arrow).

affected site in 60% of cases, followed by thoracic (19%) and cervical spine (12%). Multi-level involvement is an exceptional form of brucellar spondylitis found in only 3%

of the cases.  $^{1-5}$  In these situations, involved vertebrae are mostly contiguous. To the best of our knowledge, only five similar cases have been found in the world literature.  $^{1-5}$ 

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In all reported cases, they were men aged more than 60 years old with a history of exposure to dairy animals or ingestion of raw dairy products. The patients frequently also reported fever, night sweats and weight loss. Plain radiographs of the affected spine can show some abnormalities, but MRI of the whole spine revealed abnormal signal intensity of the discs and adjacent vertebrae involved levels. Paraspinal and epidural abscesses has been reported in some cases.<sup>5</sup>

## **Learning points**

- Multi-level involvement is an exceptional form of brucellar spondylodiscitis.
- MRI is the imaging method of choice for the diagnosis and follow-up of brucellar spondylodiscitis.
- There are many difficulties in diagnosing the aetiological agent Brucella and in differentiating its specific features from tuberculosis, particularly in endemic regions.

Competing interests None.

Patient consent Not obtained.

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Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Mrabet D, Mizouni H, Khiari H, Rekik S, Chéour E, Meddeb N, Mnif E, Mrabet AB, Srairi HS, Sellami S. Brucellar spondylodiscitis affecting non-contiguous spine levels. *BMJ Case Reports* 2011;10.1136/bcr.01.2011.3788, date of publication

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