

Cervical and lumbar diffuse *Brucella* spondylodiscitis: an uncommon case report

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ABSTRACT:

- **Introduction:** *Brucellosis* is a systemic zoonotic disease frequently involving musculoskeletal system, specifically the osteoarticular structures. Lumbar spine involvement is the most prevalent, whereas cervical spine is a rare location.
- **Case presentation:** We reported a case of a 59-year-old man with a 6-month history of lumbar and cervical back pain, restricted neck and lumbar movements, high fever and night sweats. He revealed the consumption of unpasteurized milk products. MRI showed L1–L2 intervertebral disc destruction and contrast-enhanced of the vertebral body at C5–C6 and L1–L2 levels with paraspinal involvement. *Brucella* serology and agglutination test resulted positive. Antibiotic therapy with Rifampicin, Doxycycline and Ciprofloxacin was administered. Patient achieved fever's disappearance together with a prompt amelioration of lumbar and cervical pain.
- **Discussion:** This is an uncommon case of contemporary *Brucella*'s involvement of lumbar and cervical spine. Serology, cultural test and imaging along with medical history are important to make a rapid diagnosis of brucellosis' osteoarticular complications. Standard antibiotic therapy regimen and its duration have not been reported yet.
- **Keywords:** Brucellosis, Spondylodiscitis, *Brucella* osteoarticular involvement.

INTRODUCTION

Brucellosis is one of the most common microbial zoonotic diseases worldwide and it is endemic in large part of developed and developing countries¹. Brucellosis is caused by *Brucella*, an intracellular gram-negative coccobacillus and *Brucella melitensis* is the species responsible for the majority of human infections and diseases^{1,2}.

Thousands of new cases of brucellosis are reported annually worldwide and it is now well-known that humans could acquire the infection in two main ways: consumption of contaminated dairy products (milk, butter and cheese), or by occupational contacts (e.g. veterinary and butcher)^{3,4}.

Brucellosis is a systemic infection but the musculoskeletal system is frequently interested⁵ and, in detail,

the most frequent complication of brucellosis is osteoarticular involvement, which can occur in 10% to 85% of the patients with the disease⁶. We report an uncommon and very challenging clinical case of *Brucella* spondylodiscitis involved both cervical and lumbar spine.

CASE PRESENTATION

On June 2019, a 59-year-old man with a 6-month history of lumbar and cervical back pain, restricted neck and lumbar movements, high fever (39°C) and night sweats, was admitted to the Infectious Diseases Department of the Garibaldi Nesima Hospital (Eastern Sicily).

The patient was a carpenter and reported usual consumption of unpasteurized milk products.

He was overweight and his medical history showed hypertension treated with ARB and impaired glucose tolerance treated with metformin; he had never had major surgery and he had never undergone any invasive medical procedures.

On admission, the patient was febrile (T. 38°C), blood pressure was 140/80 mmHg, heart rate was 108 beats/min, and oxygen saturation was 98% in room air. Physical examination revealed intense lumbar and cervical back pain with decreased range of motion, VAS score was 8; no neurological deficit or meningeal signs were noted.

Magnetic resonance imaging (MRI) revealed L1–L2 intervertebral disc destruction with a reduced height of the disc space and abnormal signal intensity of contiguous vertebral bodies; in T2-weighted scan, it showed contrast-enhanced of the vertebral body at C5–C6 and L1–L2 levels. In both localizations, the presence of paraspinal involvement was reported (Figure 1).

Laboratory examinations showed white blood cell count 5600/mm³; hemoglobin 11.5 g/dL; platelet count 235000/mm³; ESR 88 mm/H; CRP 8.73 mg/dL, LDH 282 UI/L.

Brucella serology using enzyme-linked immunosorbent assay was positive for IgG and negative for IgM with two agglutination tests (Wright test) positive with a titer of 1:640. Blood cultures were negative. Mantoux test and QuantiferonTB resulted also negative.

A PET/TC scan was performed showing pathological uptake in C7 vertebral body with altered uptake in L1–L2 level.

Given the clinical presentation, imaging and positive Brucella serology test, the diagnosis of multifocal brucellar spondylodiscitis was established.

Patient's conditions were discussed with the spine surgeon and with orthopedic to evaluate the need for surgical

intervention and it was decided to continue on medical treatment alone, as there was no neurological deficit. The cervical spine was immobilized with Philadelphia neck collar and the lumbar spine with an orthopedic corset.

Antibiotic therapy was started with Rifampicin (600 mg + 300 mg IV daily), Doxycycline (100 mg po twice daily) and Ciprofloxacin (500 mg po twice daily). During antibiotic therapy, patient's conditions improved rapidly with a prompt amelioration of lumbar and cervical pain and partial functional recovery. Fever disappeared with a significant reduction of inflammatory markers (after one month of therapy, ESR was 30 mm/h and CRP was 0.98 mg/dl, VAS score was 3). 6 weeks after the patient was discharged with the recommendation to continue antibiotics, which duration would have been established on the basis of the result of clinical data and imaging. He is now followed up as out patient with no evidence of disease recurrence.

DISCUSSION

Brucella-related musculoskeletal involvement is the most common manifestation of systemic brucellosis reported in 10–80% of diseases; spondylitis, spondylodiscitis and discitis are the main manifestations^{7,8}.

Kulowski and Vinke⁹ reported the first proved case of *Brucella melitensis* involvement of the human spine and up until now, there have been reported several cases about spinal brucellar infection¹⁰.

Lumbar (60%), sacral (19%) and cervical (12%) vertebrae were the most common affected sites^{12,13} but compared with lumbar spondylitis, the cervical involvement is associated with a higher incidence of paravertebral or epidural masses, neurological complications, and an overall worse prognosis^{13,14}. To validate this, Pina et al¹⁵ presented a rare case of *Brucella* spondylodiscitis complicated with an epidural abscess in the cervical spine whereas



Figure 1. Cervical and lumbar involvement in MRI.

Hantzidis et al¹⁶ reported a case of a 65-year-old farmer who presented with Brucella-related cervical spondylitis with spinal cord compression. There are only a few cases showing the involvement of cervical spine in Brucella infections and it is very rare to show the contemporary Brucella's involvement of lumbar and cervical spine. A case similar to ours was reported by Zormpala et al¹³ showing a case of Brucella spondylitis involving both the cervical and lumbar spine with a delayed diagnosis. Serological and cultural test, imaging methods together with medical history are important to make a prompt diagnosis of spinal brucellosis¹⁶. Microbial isolation is the gold standard for the definite diagnosis; however, as showed in our case, blood cultures could be negative (especially in old infections) and culture from bone marrow are often too invasive. Therefore, a presumptive diagnosis is often made with serological tests, imaging and a well-collected medical history. Nevertheless, it's important to rule out other causes of spondylitis such as tuberculosis, tumoral causes and complications secondary to trauma^{17,18}.

Because of the negativity of Mantoux and QuantiferonTB we were able to rule out tuberculosis, also, the patient had no trauma history and the imaging along with TC/PET weren't suggestive for tumoral diseases. Treatment options for Brucella osteoarticular involvement include antibiotic therapy and, when it failed or for patients with neurological deficits, surgical intervention^{18,19}. A standard therapy for osteoarticular brucellosis has not been reported yet; however, it's safer to prevent relapses using a triple regimen containing Doxycycline and Rifampicin together with a 3rd antibiotic⁵ at least for three months, even if our experience together with literature²⁰ suggest prolonged treatments over 6 months had much better outcomes and very few relapses. As a 3rd drug, we chose Ciprofloxacin due to its excellent effect against Brucella and its great penetration in bone tissue^{22,23}.

CONCLUSIONS

In patients complaining about fever and back pain, it is important to consider Brucellosis as a differential diagnosis, especially in endemic regions, together with its complications and promptly start the appropriate diagnostic pathway. In subjects with brucellar spondylitis, the whole spine should be quickly evaluated with imaging, preferably with MRI, in order to assess damage's extension and to prevent neurological complications. Antibiotic therapy is the first treatment choice and it should be administered in triple regimen and at least for three months, considering quinolones as a potential third drug class in addition to standard drug regimen consisting in Doxycycline and Rifampicin.

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FC and AM wrote the paper. AO, MG, AZ and ST gave clinical assistance to the case. ML, AP, DS, AG and MC searched literature references. BC and GN revised the paper. All authors read and approved the final manuscript.

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Not applicable.

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Written informed consent for publication of clinical details was obtained from the patient and it is contained in the patient's clinical record.

CONFLICT OF INTEREST:

The authors declare that they have no conflict interests.

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