Spinal Cord (2003) 41, 574–576

www.nature.com/sc

Case Report

Neurobrucellosis and a demonstration of its involvement in spinal roots via magnetic resonance imaging

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Objective: To present a neurobrucellosis case with spinal root involvement by means of magnetic resonance imaging (MRI).
Methods: A case of neurobrucellosis resembling Guillain–Barré syndrome is being reported. This case is unique in a way that spinal root involvement because of brucellosis was for the first time confirmed by MRI.
Setting: Spinal cord unit of a rehabilitation and care center in Ankara, Turkey.
Results: The correct diagnosis was made with cerebrospinal fluid culture. The patient showed a significant improvement with antimicrobial therapy and rehabilitation.
Conclusion: Polyradiculopathy because of neurobrucellosis may mimic neurological syndromes. Rehabilitation should also be a part of its treatment.
Spinal Cord (2003) **41,** 574–576. doi:10.1038/sj.sc.3101479

Keywords: neurobrucellosis; paraplegia; polyradiculoneuropathy; magnetic resonance imaging

Introduction

Brucellosis is a disease because of infection with Gram-negative microorganisms of the genus *Brucella*.[1] It is a zoonosis transmittable to humans. All *Brucella* infections are led by direct or indirect exposure (eg, milk or milk products and raw meat) to animals.[2] The disease starts with nonspecific symptoms such as fever, sweats, malaise, anorexia, headache, and back pain. The diagnosis has been made from a history of previous symptoms of the disease, culture of the organisms from the blood or cerebrospinal fluid, and serologic testing.[1] Long-term antimicrobial therapy has been regarded as an effective treatment strategy for brucellosis.[2] Among the most commonly involved systems because of *Brucella* are the gastrointestinal tract, the hepatobiliary system, and the skeletal system.[1] However, nervous system involvement is not common. The incidence has been reported to be between 3 and 25% of the cases of generalized brucellosis.[3] Its effect on the spinal cord may be becasue of compression of abscess and granuloma or involvement of the spinal roots. The purpose of this article was to present a neurobrucellosis case with spinal root involvement that could be confirmed by magnetic resonance imaging (MRI).

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Case report

A 21-year-old male patient was admitted to our rehabilitation center with paraplegia because of brucellosis. His medical history had started with back and left leg pain 10 weeks before his admission. He had no previous episode of fever or malaise. A progressive strength loss in his lower extremities has appeared within a week. He was then hospitalized in a neurology unit. Electromyography, which was performed 4 weeks before admission, revealed a pure motor polyradiculoneuropathy. Studies on distal nerve conduction indicated no motor or sensory abnormalities. Somatosensorial-evoked potentials of tibial and median nerve were normal as well. F-wave and H-reflex latencies were slightly prolonged. The lumbar motor-evoked potential latencies were prolonged and the amplitudes were reduced. Needle electromyography of his lower extremity muscles showed denervation. MRI demonstrated the involvement of the nerve roots. As can be seen in Figure 1A and B, sagittal and axial postcontrast T1-weighted images showed thickening of the spinal roots and diffuse enhancement along the distal cord and cauda equina. Unfortunately, the patient was misdiagnosed with Guillain–Barré syndrome and had undergone intravenous immunoglobulin therapy and plasmapheresis. Apparently, he had no benefit from these therapies and his symptoms got worse during the treatment. Therefore, new tests for a diagnostic purpose

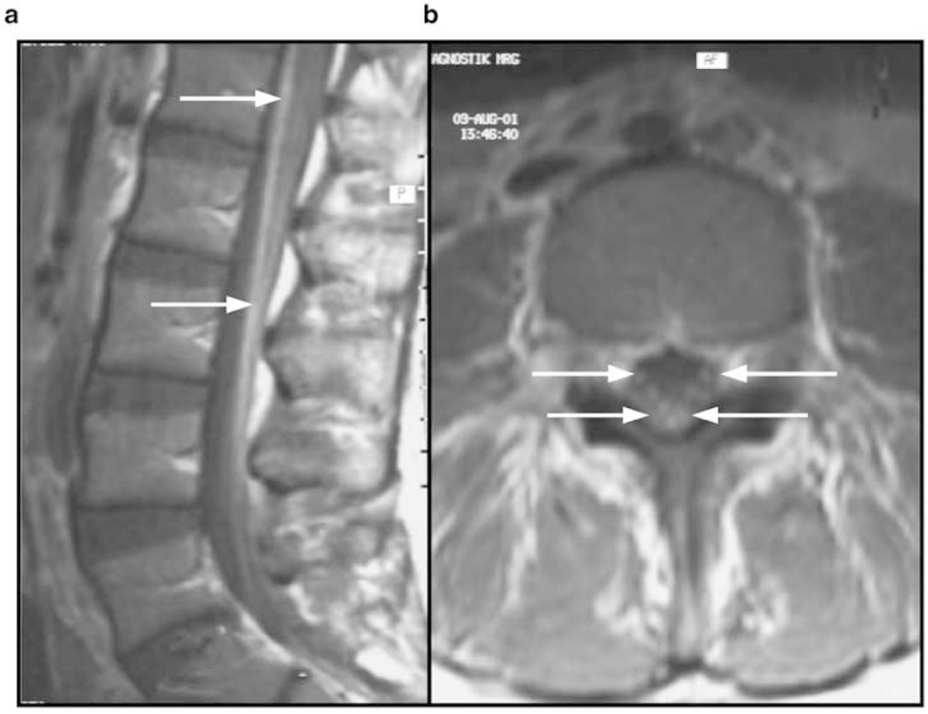

Figure 1 (**a**) Postcontrast sagittal T1-weighted image showed diffuse contrast enhancement on the distal cord and cauda equina (arrows). (**b**) Postcontrast axial T1-weighted image from the level of the fourth lumbar vertebra showed thickening and enhancement of the nerve roots (arrows). These findings indicated nonspecific inflammation involving the distal cord and the nerve roots

were performed. The Rose Bengal test yielded positive results both in blood and cerebrospinal fluid (CSF). CSF had a xanthochromic appearance with elevated protein level (319 mg/dl), lymphocytic pleocytosis (700 cells/μl, 95% lymphocytes), and low sugar (35 mg/dl). The diagnosis was changed to brucellosis upon the identification of *Brucella melitensis* in CSF culture. Antimicrobial therapy was started with ceftriaxone 4 g/day intravenously and rifampin 600 mg/day orally. Afterwards, the patient was referred to our center for rehabilitation.

On admission, the patient was suffering from strength loss in his lower extremities. The strength of his lower extremity muscles was at grade 0 with manual muscle testing except for grade 1 hip flexors and ankle plantar flexors on either side. No sensory loss was detected. He had flaccid paralysis and his muscle stretch reflexes were absent. Pathologic reflexes could not be elicited. Bulbocavernous and anal reflexes were normal and the patient had no bowel or bladder dysfunction. Routine blood and urine analysis did not reveal any pathologic results. A rehabilitation program including range of motion exercises, strengthening, and ambulation with orthotic devices was started. His medical treatment was changed to doxycycline 200 mg/day and rifampin 600 mg/day orally at the end of the 8th week. The patient has showed a significant clinical improvement. After 3 months of rehabilitation and medical therapy, he was able to walk with minimal assistance without any orthotic devices. The muscle strengths of his knee extensors and ankle plantar flexors were improved to grade 5, and the rest of the lower extremity muscles to grade 4 on each side. In the rehabilitation period his muscle stretch reflexes were unchanged. At 4 months after the first electromyography, the patient was re-examined electrophysiologically. The new electromyographic findings were similar to those of the previous examination, although his clinical improvement was almost excellent. Rehabilitation and medical therapy of the patient has yet to be terminated.

Discussion

Brucellosis is a systemic infection in which any organ or system of the body can be involved. Nervous system complications are rare, but cases of meningitis,[4] abscess development,[5] granulomas,[6,7] and spondylitis[8,9] have been reported in the literature. In endemic areas any suspicious nervous system symptom should be investigated for neurobrucellosis as it may easily be misdiagnosed. Spinal granuloma or abscess because of brucellosis may cause an upper motor neuron-type lesion, whereas brucellar spinal root involvement may cause a lower type of motor neuron lesion. In this regard, our case is a lower motor neuron type lesion with its nerve root involvement. A Guillain–Barré like syndrome with symmetrical polyradiculopathy and no sensory loss has been reported by Bahemuka *et al*[10] and five cases of polyradiculoneuropathy have been previously reported by Kochar *et al*.[11] Besides, Shakir

et al[12] reported 19 cases of neurobrucellosis, six of which were in the form of proximal polyradiculoneuropathy. Similar to our case, no clinical sensory impairment was noted and no abnormality in sensory nerve conduction was detected in any of Shakir's cases. On the other hand, Kochar reported mild sensory changes in his series of five patients. Unlike the findings of Shakir and Kochar, distal motor latencies were normal in our case. The rest of the clinical and laboratory findings of our patient were, in general, consistent with the findings of both studies. However, the diagnosis of polyradiculoneuropathy was made with clinical and electromyographic examinations in these studies. This study presented via MRI a case that brucellosis can involve or demonstrate its effects in the roots of the spinal cord. To the best of our knowledge, this is the first case in medical literature written in the English language where spinal root involvement because of brucellosis could be confirmed by an imaging study.

Scholars have argued that the effect of brucellosis on the nervous system can be because of the direct effect of bacilli, cytokines, or endotoxins on peripheral nerves, spinal cord, meninges, and brain.[11] Although the exact pathology is not totally resolved, a demyelinating lesion is possible to account for some of the features.[12] Our patient showed marked clinical improvement in a relatively short time period despite persisting electrophysiological abnormalities. Kochar also reported a similar case. The contradiction between the clinical progress and electrophysiological status of the patients implies a peripheral demyelinating process in these cases.

Brucellosis has been primarily treated with antibiotics. Nevertheless, rehabilitation should be a part of its treatment when the nervous system is involved. It is essential not only for improving the neurological status of the patient but also for preventing complications such as contracture, pressure sores, and vascular problems.

In conclusion, it should be kept in mind that polyradiculoneuropathy because of brucellosis may resemble a variety of neurological syndromes. Besides, rehabilitation is of great importance in its treatment.

Acknowledgements

We thank Dr H Coskun for his support for review of the manuscript and for insightful comments.

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