Original Article

Isolated Cervical Spondylitis due to Brucellosis

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Abstract

Introduction: Brucella is a gram-negative coccobacillus which can affect humans as an accidental host and cause brucellosis; also known as "Malta fever". Brucellosis could manifest in several ways and can attack all the organs. Musculoskeletal complaints considered the most common complication in patients. The Involvement of the lumbar region is much more frequent than the cervical or thoracic region. This article proceeds to a case with isolated cervical brucellosis. **Case Presentation:** A 39-year-old Iranian woman presented to an infectious disease clinic with sub-acute neck pain, bilateral paresthesia of upper limb and occasional but profuse night sweats. Two weeks earlier she was prescribed NSAIDs by a neurologist which resulted in worsening of her symptoms. She was otherwise healthy. She mentioned a trip to India three months ago where she consumed local-unpasteurized dairy products. She had a history of recent weight loss of 3 kilograms. On neurologic examination, the neck range of motion was restricted and tenderness on the lower cervical spine was detected. Distal and proximal extremity forces were reduced bilaterally (4/5) with decreased deep tendon reflexes of biceps and sensory deficit which was more significant on the palmar aspect of fingers 1-3, consistent with C7 dermatome. blood cell count was normal, (ESR): 20 mm/h, C-reactive protein (CRP): 13 mg/L, wright: 1/1280, 2ME: 1/640. Blood cultures came back negative. Spinal MRI scan revealed an abnormal signal intensity of C4 to C6 vertebrae and a decrease in height of C5/C6 intervertebral disc suggestive of spondylodiscitis plus anterior wedging of C5 with endplate irregularity. The patient to start on outpatient treatment with Gentamicin plus Doxycycline plus Rifampin. After 2 weeks of treatment initiation, spinal tenderness decreased significantly and the neck range of motion restored. CRP and ESR decline to normal values (2 mg/L and 5 mm/h respectively). The patient immigrated to another country and went out of reach, so follow-up imaging had not been performed, but follow-up MRI advised to be done at the end of the treatment course to evaluate regimen efficacy and probable need to treatment extension. **Conclusions:** It should be kept in mind that brucellosis may present with a painful and stiff neck in patients who reside in endemic regions or have traveled to those countries.

Keywords: spondylodiscitis, brucellosis

Introduction

Brucella is a gram-negative coccobacillus which can affect humans as an accidental host and cause brucellosis; also known as "Malta fever" [1]. The disease considered as high-prevalent in Asia, Middle East, Africa, South, and Central America. Brucellosis could manifest in several ways and can attack all the organs. Besides constitutional symptoms, it can be presented with musculoskeletal complaints, testicular swelling, endocarditis, neurologic disorders, liver and/or spleen enlargement, hematologic disorder, depression, etc. [1-4]. Regarding this heterogeneity of signs and symptoms, it can easily be mistaken with more prevalent diseases like disk herniation, tuberculosis, malaria and other febrile conditions [5]. Musculoskeletal complaints considered the most common complication in patients [1, 4-6]. It can manifest as sacroiliitis, bursitis, tenosynovitis, osteomyelitis, spondylitis, discitis, spondylodiscitis, and paravertebral abscess [7, 8]. Peripheral nervous system involvement can overlap in presentation, includes neuropathy, radiculopathy and ascending myelitis [1]. The Involvement of the lumbar region is much more frequent than the cervical or thoracic region [5]. This article proceeds to a case with isolated cervical brucellosis.

Case Presentation

A 39-year-old Iranian woman presented to an infectious disease clinic with sub-acute neck pain, bilateral paresthesia of upper limb and occasional but profuse night sweats. Two weeks earlier she was prescribed NSAIDs by a neurologist which resulted in worsening of her symptoms. She was otherwise healthy and did not use any other medication or

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illicit drugs. She mentioned occasional alcohol consumption and a trip to India three months ago where she consumed local-unpasteurized dairy products. She had a normal appetite, meanwhile a history of recent weight loss of 3 kilograms. On examination vital signs were stable. No sign of anemia, abnormal lung or heart sounds, hepatomegaly, splenomegaly, and lymphadenopathy were detected. On neurologic examination, the neck range of motion was restricted and tenderness on the lower cervical spine was detected. Distal and proximal extremity forces were reduced bilaterally (4/5) with decreased deep tendon reflexes of biceps and sensory deficit which was more significant on the palmar aspect of fingers 1-3, consistent with C7 dermatome. On blood count, white cell count: 10.800/mm^3 (PMN: 65%, LMN: 30%), red cell count: 4100/mm^3, hemoglobin: 12.1 g/dl, platelet count: 230.000/mm^3, erythrocyte sedimentation rate (ESR): 20 mm/h, C-reactive protein (CRP): 13 mg/L, wright: 1/1280, 2ME: 1/640. Blood cultures came back negative. Spinal MRI scan (Figure 1) revealed an abnormal signal intensity of C4 to C6 vertebrae and a decrease in height of C5/C6 intervertebral disc suggestive of spondylodiscitis plus anterior wedging of C5 with endplate irregularity. No other vertebra was involved. Abdominal ultrasound findings were non-significant. The patient to start on outpatient treatment with Gentamicin (5 mg/kg/day for 14 days) plus Doxycycline (100 mg BID for 12 weeks) plus Rifampin (600 mg/day for 12 weeks). In the first week of treatment, her symptoms deteriorated and upper extremity forces decreased to 3/5. She started on Acetaminophen (3000 mg/day) and her symptoms begin to shrink. After 2 weeks of treatment initiation, spinal tenderness decreased significantly and the neck range of motion restored. CRP and ESR decline to normal values (2 mg/L and 5 mm/h respectively). The patient immigrated to another country and went out of reach, so follow-up imaging had not been performed, but follow-up MRI advised to be done at the end of the treatment course to evaluate regimen efficacy and probable need to treatment extension. The patient obtained a plain radiograph in an outpatient clinic 4 weeks following treatment initiation (Figure 2).

Discussion

Brucella is a facultative intracellular pathogen. Its transmission can occur mainly through the consumption of unpasteurized dairy products. Both genders contract the disease equally, but depending on occupational status, it is probable that high risk exposures be more frequent in males (working in a slaughterhouse or as a shepherd) [5]. B. melitensis is in charge of most osteoarticular complications specifically in high-prevalence areas (Mediterranean Sabin, Middle East, South, and Central America , Central Africa), whereas B.abortus and B.suis commonly cause these form of complications in low-prevalence areas [5, 8]. This patient recently had traveled to India and due to previous epidemiological studies, we suspected B.melitensis [9]. Unlike our patient, who had a convincing history of how she was infected, a precise transmission route usually cannot recognize through a patient‘s history in about 40% of cases [10, 11]. Thus in endemic areas, health care providers need a high index of suspicion when a patient complains of unspecific flu-like symptoms like fever, fatigue, and myalgia. It is impossible to distinguish Brucella strain according to clinical manifestations [12]. Culture of body fluids/or tissue is considered as the gold standard of diagnosis and its accuracy declines as the disease gets more chronic [5]. Regarding high rates of culture failure to establish the diagnosis, practically, immunological methods (Wright test, 2-mercaptoethanol test, complement fixation test, coombs test,Enzyme-linked immunosorbent assay) provide more benefits in diagnosis [5, 13]. If any doubts arise while interpretation of results, it is advised to perform these modalities in multiple times with proper intervals to gain more reliable results [5].

According to several studies, among general manifestations of brucellosis, sweats (34%), headaches (28%) and fever (26%) are the most common complaints [4, 14]. About musculoskeletal symptoms, arthralgia (37%), arthritis (26%) and back pain (23%) are the majority of complaints [4, 14]. Osteoarticular involvement occurs at least in 27 percent of patients [14, 15]. The osteoarticular complications are not specific to the Brucella species which affected the patient, moreover, they probably is determined by genetic factors such as presence of HLA-B39 [12]. Vertebral involvement accounts for about 10% of these manifestations [8, 16]. It presents as local spinal tenderness in 78-97% of patients [17]. Some risk factors for spinal involvement in brucellosis are diabetes mellitus, alcohol consumption, dialysis associated with chronic renal failure [13, 18]. Brucellosis tends to involve the spine in the following order: lumbar (60%), thoracic (19%) and cervical (12%) region [5, 7, 13, 16]. Interestingly, spine involvement might occur in multiple levels of the spinal column [7, 16, 18]. 5-36% of patients with brucellar spondylitis found to have multilevel spinal lesions [16]. Spinal brucellosis can occur locally and diffusely [7, 19]. In local form, only the anterior portion of the end plate is affected. Any involvement beyond this territory considered as a diffuse form where neighboring discs and spines might be affected [18]. Inversely, epidural abscesses mostly arise from the posterior part of epidural space [6]. MRI considered the modality of choice for spinal brucellosis [11]. It is about 96% sensitive and can identify spine inflammation within the first month of its initiation [6, 17]. It also can differentiate other causes of spondylitis such as tuberculosis (Table 1), pyogenic spondylitis, metastasis, and degenerative disorders of the spine [8, 16]. Plain radiographs, CT scan, and scintigraphy could be used in specific situations but in early stages of spondylitis, radiographs and even CT scan might miss the diagnosis [11]. scintigraphy is sensitive in initial stages but lacks specificity [8, 18]. Some studies use MRI as a follow-up tool. Oddly, spinal involvement in brucellosis can occur through direct (septic) and indirect (immune-mediated) form. Although no standard antibiotic regiment proposed to treat brucellar spondylitis, it is generally accepted that the duration of treatment should be extended when complications such as spondylodiscitis or paravertebral abscess are present [18, 20]. Ioannou et al. proposed that the minimum duration of

treatment should be 24 weeks providing that radiologic resolution achieved. If not, treatment duration should be extended even to 72 weeks and by this guideline, none of their patients experienced relapse (with triple therapy) [21]. Surgical or CT-guided drainage is necessary if the proper response is not achieved and disabling complications like nerve compression, cauda equina syndrome or paravertebral abscess occur [11].

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Conflict of interest:

The authors declared that they had no conflict of interest.

Ethical Approval:

This study was approved by ethical committee of Mashhad University of Medical Sciences.

Figures and Tables

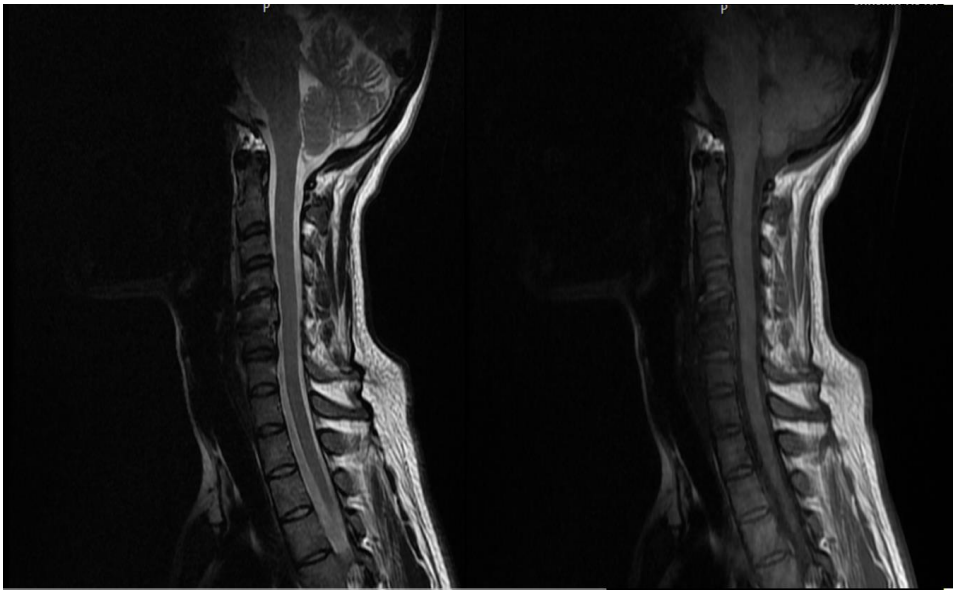

Figure 1: T1 and T2 MRI findings

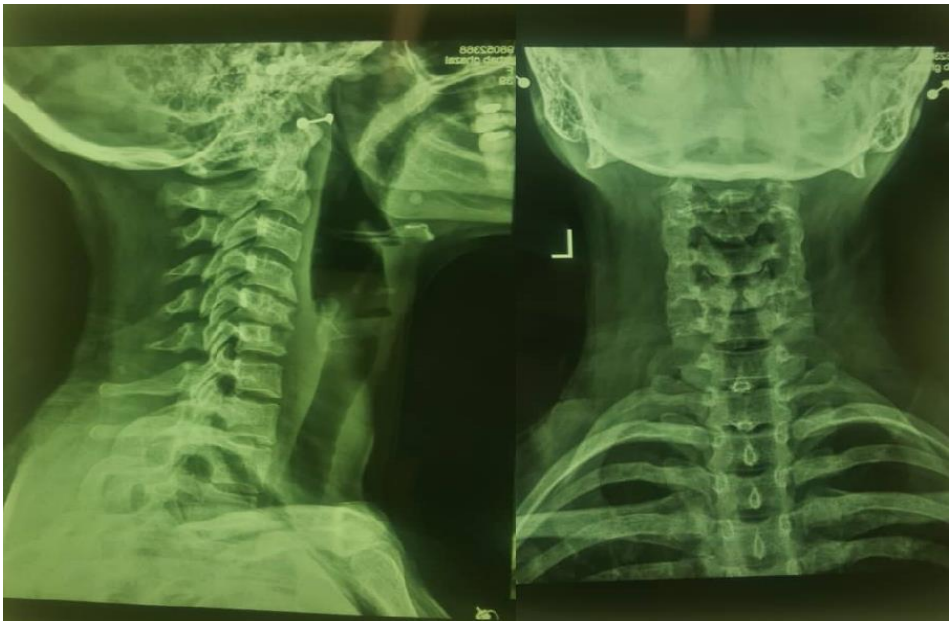

Figure 2: plain radiograph findings after 4 weeks of treatment

Table 1: MRI findings of Brucellar vs. Tubercular spondylodiscitis.

		Intervertebral disc involvement	Paravertebral soft-tissue involvement	spine
Brucellosis	acute	T_1W hypo-intensity + T_2W hyper-intensity	Small	anterior
	chronic	$T_{1\&2}$ W heterogeneous intensity		
Tuberculosis		No abnormal signal	Significant	posterior

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