Case Report

Myositis as the first presentation of acute brucellosis - A case report

Abdulaziz Alfaifi[1], Yasir Al Tuwairqi[2], Naif Alomairi[2], Awatif Edrees[2], Ali Al Ahmari[3], Ibrahim Masoodi[2]

[1]Department of Internal Medicine, King Abdul Aziz Specialist Hospital, Taif, Kingdom of Saudi Arabia, [2]Department of Internal Medicine, College of Medicine, Taif University, Taif, Kingdom of Saudi Arabia, [3]Department of Gastroenterology, Al Hada Military Hospital, Taif, Kingdom of Saudi Arabia

Correspondence to: Ibrahim Masoodi, E-mail: ibrahimmasoodi@yahoo.co.in

Received: April 11, 2018; **Accepted:** October 16, 2018

ABSTRACT

The clinical scenario of a young male is presented in this report who presented with severe pain and swelling of both lower limbs of 3-day duration. On evaluation, he proved to have myositis and further evaluation unraveled *Brucellosis*. The index case improved with conventional *Brucella melitensis* treatment confirming the cause and effect relationship. The *Brucella* infection is a male predominant disease and frequently presents with typical symptoms of fever, fatigue, etc., and the diagnosis often is straightforward. Nevertheless, neurobrucellosis is uncommon, developing in <5% of patients with *Brucella* infection and producing diverse neurological syndrome. There are very few reports of Myositis caused by Brucella described in the literature. The heaviest disease burden of *B. melitensis* lies in countries of the Mediterranean basin and Arabian Peninsula. However, the disease is not uncommon in India, Mexico, and Central America. Hence, clinicians need to be aware of this clinical entity all over the globe. Myositis as the first presentation of acute brucellosis is presented in this report.

KEY WORDS: Acute Brucellosis; Myositis; Swollen Limbs

INTRODUCTION

Brucellosis (Mediterranean fever, undulant fever) is the most common zoonotic infection caused by the bacterial genus *Brucella*. These organisms are small aerobic intracellular coccobacilli, which localize in the reproductive organs of the host animals and the humans contract infection by ingestion through infected food products, direct contact with an infected animal, or inhalation of aerosols. Even though humans are accidental hosts, brucellosis continues to be a major public health concern worldwide. Brucellosis is estimated to cause more than 500,000 infections per year worldwide. Its geographic distribution is limited by effective public and animal health programs, and the prevalence of the disease varies widely from country to country.[1] Overall, the frequency of brucellosis is higher in more agrarian societies and in places where handling of animal products and dairy products is less stringent. Brucellosis in humans is a multisystem disease that may present with a broad spectrum of clinical manifestations. The central nervous system involvement in brucellosis sometimes can cause demyelinating syndromes. Brucellosis is endemic in Saudi Arabia and presents with classical symptoms of fever, fatigue, and malaise. In a study from northern Saudi Arabia, authors observed that for unknown reasons majority of brucellosis cases (60%) were in the age range of 13–40 years, whereas 21% occurred in those younger than 13 years, 16% in those aged 40–60 years, and 2.5% in those older than 60 years.[2] *Brucella* presenting as myositis is presented in this report.

Access this article online	
Website: http://www.ijmsph.com	Quick Response code
DOI: 10.5455/ijmsph.2019.1028516102018	

CASE REPORT

A 17-year-old male presented with a history of pain and swelling in both lower limbs of 1-week duration following low-grade fever 10-day duration. Pain was severe and required round the clock analgesics. He denied any high-risk

behavior, trauma, or long travel. Fever had been low grade with no diurnal variation and was associated with occasional chills. The examination of the patient revealed no pallor or lymphadenopathy. The pulse was 74 beats/minute regular, blood pressure was 120/80 mm of mercury, and jugular venous pressure was normal. The temperature documented in the hospital was 38°C. The abdominal examination showed mild splenomegaly. The chest and cardiovascular examination were normal. Both lower limbs were uniformly swollen and there was non-pitting edema as shown in Figure 1. However, power was grade 5/5 in all four limbs. The detailed neurological examination was normal. Septic screen was sent soon after the admission and the laboratory reports in the evening as shown in Table 1 demonstrated leukocytosis (14 × 10^3 raised erythrocyte sedimentation rate [ESR] = 60 mm 1st h). The X-ray chest was normal. An initial impression of deep venous thrombosis was made in Emergency room of the hospital ER; however, compression ultrasound did not reveal any deep venous thrombosis. Then, MRI of both limbs was done which revealed bilateral nearly symmetrical bulky edematous posterior leg muscle group with abnormal signal in the form of increased flair and T2 W1 signal intensity. There was associated hyperintense fascia surrounding muscle on FLAIR and T2 W1. The myofascial abnormalities were more prominent in the right side and did not exceed above knee level bilaterally. No subcutaneous abnormalities were noted and an impression of inflammatory myopathy with fasciitis was made [Figure 2]. Electromyography of the calf muscles showed low amplitude, polyphasic, and myopathic units, the interference pattern indicated myositis.

Brucella serology was positive and 1:260 titers. The patient was started on conventional *Brucella* treatment, injection streptomycin 0.75 gm intramuscular once daily and tablet doxycycline 100 mg twice daily. He became a febrile and pain in the limbs decreased and 4 weeks later the swelling also reduced [Figure 3]. The blood culture grew *Brucella melitensis* and standard anti-*Brucella* regimen was continued for a total of 6 weeks. On repeat questioning, the patient admitted ingestion of raw camel milk few weeks before the current presentation.

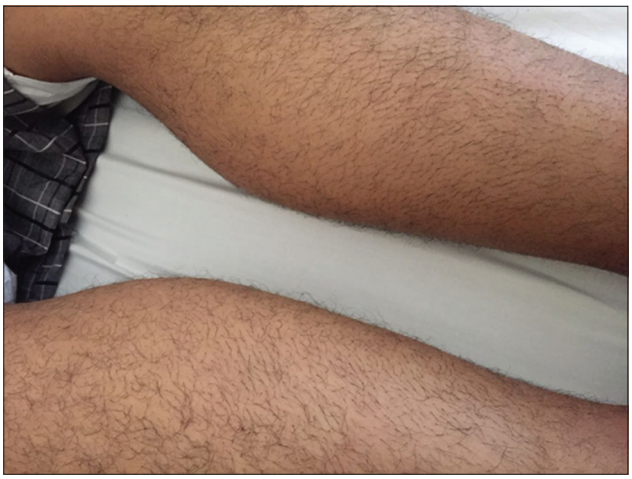

Figure 1: Lower limbs - uniformly swollen with non-pitting edema

DISCUSSION

The index case was brought to emergency with bilateral painful lower limbs. In the beginning of his hospital course, brucellosis was difficult to predict as the cause of pain and swollen limbs. Even though he had no risk factors for deep vein thrombosis, clinical scenario demanded that this condition be ruled out. As a part of workup for fever of unknown origin. Brucella serology was sent as Saudi Arabia continues to be an endemic zone of Brucellosis.[2] It was only after the results of Brucella serology, treatment for the condition was started to which he showed dramatic response with decline in his fever, and over a period of few weeks, swelling of limbs also subsided. A high degree of suspicion in the diagnosis of brucellar myositis is essential to reduce the delay for the treatment. There is a paucity of data regarding

Table 1: The investigations before and after treatment

Test	Result		Normal range
	Pre-treatment (on presentation)	Post-treatment (after 6 weeks)	
Hemoglobin	13.4	14.3	12.2–15.3 g/dl
White blood cell	14	6.2	6-16×109/l
Platelet	21,000	197	150–450×109/l
ESR	60 mm 1st h	22 mm 1st h	
Total bilirubin	1.2	1.0	0.8–1 mg/dl
Direct bilirubin	0.8	0.8	0–0.6 µmol/L
AST	45	30	5–30 U/L
ALT	56	29	5–30 U/L
ALP	100	100	50–100 U/L
GTT	46	30	7–30 IU/l
Albumin	3.9	39	38–54 g/l
Total proteins	4.2	4.5	
INR	1.1	1.1	0.8–1.2
Urea	42	40	
Creatinine	1.0	1.0	
Na/K	141/3.8	142/3.6	
Serum glucose	100	102	65–110 mg/dl
CPK	2500	112	
Brucella serology	1:320	1:112	Strongly positive
LDH	285 U/L	100U/L	50–248 U/L

AST: Aspartate transaminase, ALT: Alanine transaminase, GTT: Glucose tolerance test, CPK: Creatine phosphokinase, LDH: Lactate dehydrogenase, ESR: Erythrocyte sedimentation rate, INR: International normalized ratio

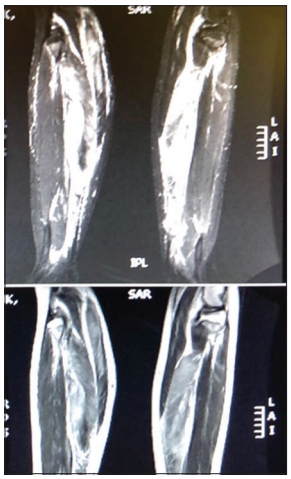

Figure 2: Magnetic resonance imaging of lower limbs - bilateral nearly symmetrical bulky edematous posterior leg muscle group

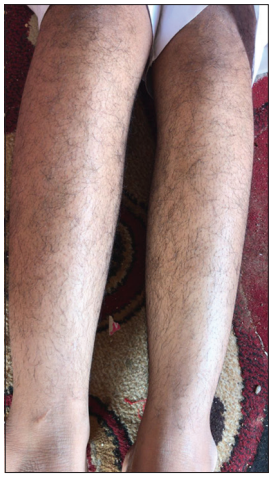

Figure 3: Reduction in swelling after 4 weeks

myositis due to brucellosis in the English literature, Aygul *et al*.[3] reported myositis in a young male who responded to conventional *Brucella* treatment like the index case. Another case of brucellar myositis was reported by Bofill *et al*.[4] The diagnosis in the index case was established by high muscle enzymes, electromyography, and positive serology and positive blood culture. Although we could not show the pathology by muscle biopsy, deposited humoral antibodies in the muscle fiber likely cause myositis or quite possible the index case had swollen limbs due to capillary leak syndrome which has been reported in the *Brucella* cases by Karsen *et al*.[5] In animal models, osteoarthritis, necrosis, periarticular soft tissue inflammation, and substantial Brucella burdens were observed by Skyberg *et al*.[6] In the above study, the authors demonstrated elevated IL-1 β, IL-6, etc., in joint homogenates. Oral rifampicin was effective in clearing infection and halting further progression of focal inflammation from infected animal model of brucellosis. In another report, polymyositis-like syndrome complicated by rhabdomyolysis secondary to brucellosis highlighting protean manifestation of acute brucellosis was reported by Kushal *et al*.[7] Progressive muscle pain, with tenderness, swelling, and diffuse, asymmetric weakness in a 16-year-old boy with muscle biopsy evidence of inflammatory granulomatous myositis was reported by Suleiman *et al*.[8] Nevertheless, the index case had normal power and never had weakness of his limbs. No muscle biopsy was done in the index case as he had high titers of *Brucella* and dramatic response to treatment. Other clues to the diagnosis in the index case were high ESR and splenomegaly. In a study from Turkey, data on 52 patients with brucellosis showed splenomegaly to be constant clinical finding followed by hepatomegaly.[9]

The antibiotic treatment remains the cornerstone modality in the management in a given case of brucellosis. The use of doxycycline and an aminoglycoside regimen seems a better strategy in selected patients. In endemic areas, the clinical suspicion needs to be very high. Atypical presentations even when serology results are equivocal the clinical suspicion of acute brucellosis should be kept in mind while evaluating a case of fever. A 26-year-old shepherd presenting with fever, ataxia, and dysarthria and weight loss was proved to have brucellosis by blood culture. He had low standard tube agglutination test (STAT) and 2-mercaptoethanol (2-ME) titers 1:80 and 1:40, respectively, quite low to suspect a diagnosis of brucellosis. Authors, in this report, have highlighted that even with low titers brucellosis should be considered in endemic areas.[10]

CONCLUSION

In endemic areas, clinicians should consider brucellosis in any unusual presentation involving multiple organ systems, even if serology is inconclusive. The current case highlights that it is important to consider brucellosis in the differential diagnosis of a patient who presents with neuromuscular findings.

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How to cite this article: Alfaifi A, Al Tuwairqi Y, Alomairi N, Edrees A, Al Ahmari A, Masoodi I. Myositis as the first presentation of acute brucellosis - A case report. Int J Med Sci Public Health 2019;8(1):101-104.

Source of Support: Nil, **Conflict of Interest:** None declared.