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Polymyositis-like syndrome with rhabdomyolysis in association with brucellosis

Kushal Naha, Suman Karanth, Sowjanya Dasari*, Mukhyaprana Prabhu

Department of Medicine, Kasturba Hospital, Manipal-576 104, Manipal University, Karnataka, India

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ABSTRACT

Diffuse myositis with progression to rhabdomyolysis has been reported in association with wide range of viral infections. We report a case of polymyositis-like syndrome complicated by rhabdomyolysis secondary to brucellosis. This case report thus contributes yet another atypical presentation to a disease already infamous for its protean manifestations.

1. Introduction

Diffuse myalgias are known to be associated with a wide spectrum of viral infections including Coxsackie virus, influenza A and B, Epstein-Barr virus, herpes simplex virus, cytomegalovirus and human immunodeficiency virus. Viral myositis per se is a severe form of muscle involvement, frequently resulting in rhabdomyolysis and acute tubular necrosis. By comparison, bacterial diseases tend to produce focal myositis, in the form of pyomyositis and spontaneous gangrenous myositis; this is due to direct invasion of the muscle by the organism. Leptospirosis and malaria can also produce diffuse myalgias although it is rarely severe enough to produce rhabdomyolysis.

2. Case report

A 35-year-old agriculturist with no premorbid illnesses, presented with fever since the past ten days. The fever was of a moderate grade, associated with chills, and intermittent in nature. He also complained of diffuse myalgias, involving his entire body. General physical examination revealed tachycardia and a mild fever. There was no significant lymphadenopathy. Neurological examination was essentially normal. Preliminary investigations revealed monocytosis (TLC: 9700, 12% monocytes), and slightly elevated creatine kinase levels (165 U/L). Pending microbiological reports, the patient was empirically treated with ceftriaxone (2 g IV q24h). By the third day of hospital stay, the patient began to complain of increasingly severe pain in his arms and legs with difficulty in getting up from a squatting position. Neurological re-evaluation confirmed the presence of proximal weakness involving both lower limbs with a grade of 2/5, as well as weakness of neck flexion. In addition, deep palpation of his muscles elicited intense tenderness. Serial monitoring of serum creatine kinase showed rapidly rising levels, peaking by day eight of hospital stay at 74890 U/L. Iso-enzyme analysis of creatine kinase yielded a pattern consistent with skeletal muscle origin (CK-MB fraction less than 1%). Aspartate aminotransferase levels were also elevated (1769 U/L), and urine tested positive for myoglobin. The overall picture was suggestive of a polymyositis-like syndrome with rhabdomyolysis. Urinary alkalinisation with parenteral sodium bicarbonate and hydration with intravenous fluids was immediately initiated to prevent the development of acute tubular necrosis. Subsequently, the patient also reported persistent difficulty in swallowing both liquids and solids. A nasogastric tube was therefore inserted to maintain adequate nutrition and forestall aspiration.

In consultation with neurologists, a nerve conduction study was done and ruled out acute inflammatory demyelinating polyneuropathy. Electromyography was performed next and demonstrated early recruitment (Figure 1), suggestive of polymyositis. Tests for anti-nuclear antibodies were negative; thyroid function tests were normal. At this point, the patient tested positive for brucellosis by the serum agglutination test at titers of 1:640. Retrospective questioning confirmed frequent occupational contact with cattle and unprocessed dairy products. Blood cultures by BacTec technique drawn at

*Corresponding author: Sowjanya Dasari, Department of Medicine, Kasturba Hospital, Manipal- 576 104 Karnataka, India.
E-mail: dasaris2001@yahoo.com
Tel: 91-0820-2922236

presentation, remained sterile up to four weeks.

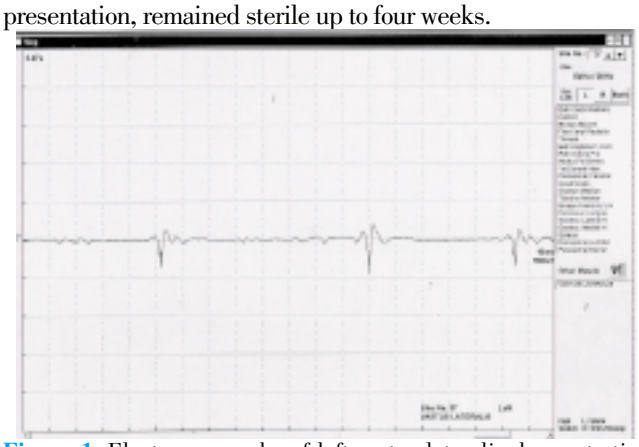

Figure 1. Electromyography of left vastus lateralis demonstrating early recruitment, suggestive of po-lymyositis.

The patient was treated with rifampicin (600 mg PO OD) and doxycycline (100 mg PO BD) for a period of six weeks. He was also administered with a short course of oral prednisolone at 1 mg/kg bodyweight, tapering over a period of three weeks. Creatine kinase returned to normal (77 U/L). Muscle tenderness, motor weakness and dysphagia resolved completely. During regular follow up till two years after the incident, the patient remained asymptomatic with no recurrences.

3. Discussion

Brucellosis is a chronic granulomatous disease that can involve virtually any organ-system of the body resulting in a protean range of clinical manifestations. It is caused by members of the *Brucella* family including *Brucella melitensis, Brucella abortus, Brucella suis* and *Brucella canis*. These facultative intracellular, aerobic, gram-negative cocco-bacilli belong to the α-2 subdivision of proteobacteria.

Brucellosis has been encountered globally, and has been reported from developed as well as developing countries, although the greater burden is still borne by poorer countries in West Asia and South East Asia. Limited awareness about the disease and lack of adequate facilities for isolation of the organism combined with the inherent tendency of brucellosis to masquerade as other diseases, have resulted in massive under-reporting of the disease undermining efforts to accurately estimate its prevalence and incidence. Nevertheless, the annual incidence of brucellosis is estimated at close to 500 000 cases[1], making it one of the commonest zoonoses worldwide.

Although brucellosis is known to present with systemic involvement ranging from osteo-articular disease to neuro-brucellosis, direct involvement of the muscular system is rare in brucellosis. A review of 100 patients by Pappas *et al*[2], did not include any patients with myositis. A larger Indian study involving 175 cases by Kochar *et al*[3], again did not report any cases of myositis secondary to brucellosis. A summary of 792 cases from several studies by Mantur *et al*[4] failed to document any cases of myositis attributable to brucellosis. A Turkish study by Buzgan *et al*[5] reported myalgias in 36.1% of 1 028 individuals with brucellosis; once again, none of these patients had evidence of myositis and/or rhabdomyolysis. A review by Calik *et al*[6] of 4 204 patients with brucellosis from three national databases of Turkey over a period of 20 years revealed only one case of myositis. Presumably, this was a case reported by Celik *et al*[7] of a 25 year old individual with culture positive brucellosis presenting with acute onset myositis[8].

Diffuse myositis resulting from infectious etiology is presumed to be immune-mediated, although definitive evidence is lacking. In the absence of positive blood cultures, it is reasonable to assume that a similar mechanism might explain the pathogenesis of myositis in our patient. Even more interestingly, this patient displayed many features classically seen in polymyositis including isolated proximal limb weakness and neck muscle weakness, and possible involvement of pharyngeal and esophageal musculature, along with a compatible electromyogram. Indeed the only evidence against our patient having a true polymyositis with incidental Brucella infection was the rapid and enduring response to a short course of oral glucocorticoids, and the negative test for anti-nuclear antibodies.

To the best of our knowledge, such a polymyositis-like pattern has not been described previously in medical literature. While the case described by Celik *et al*[7] was similar in many aspects to our own, including the massive elevation of serum creatine kinase and the electromyographic evidence of a myopathy, other features of polymyositis such as involvement of neck flexors and pharyngeal musculature were conspicuously absent. Another case report by Kojan *et al*[9] with similar findings in a 16 year old male, supported by a muscle biopsy showing an inflammatory granulomatous myositis also did not display such features of neck and visceral muscle involvement.

In summary, this case represents a unique and previously unreported manifestation of brucellosis. It also emphasizes the need for physicians working in endemic areas to maintain a high index of suspicion for brucellosis in any patient presenting with fever, regardless of the associated symptomatology.

Conflict of interest statement

We declare that we have no conflict of interest.

References

[1] Pappas G, Papadimitriou P, Akritidis N, Christou L, Tsianos EV. The new global map of human brucellosis. *Lancet Infect Dis* 2006; **6**: 91-99.
[2] Pappas G, Akritidis N, Bosilkovski M, Tsianos E. Brucellosis. *N Engl J Med* 2005; **352**: 2325-2336.
[3] Kochar DK, Gupta BK, Gupta A, Kalla A, Nayak KC, Purohit SK. Hospital-based case series of 175 cases of serologically confirmed brucellosis in Bikaner. *J Assoc Physicians India* 2007; **55**: 271-275.
[4] Mantur BG, Amarnath SK. Brucellosis in India - a review. *J Biosci* 2008; **33**: 539-547.
[5] Buzgan T, Karahocagil MK, Irmak H, Baran AI, Karsen H, Evirgen O, et al. Clinical manifestations and complications in 1028 cases of brucellosis: a retrospective evaluation and review of the literature. *Int J Infect Dis* 2010; **14**: e469-e478.
[6] Calik S, Gokengin AD. Human brucellosis in Turkey: a review of the literature between 1990 and 2009. *Turk J Med Sci* 2011; **41**(3): 549-555.
[7] Celik AD, Celik Y, Yulugkural Z, Balci K, Utku U. Acute onset myositis associated with brucellosis, quite a rare diagnosis. *Inter Med* 2008; **47**: 2091-2093.
[8] Akhtardanesh B, Ghanbarpour R, Babaei H, Nazeri M. Serological evidences of canine brucellosis as a new emerging disease in Iran. *Asian Pac J Dis* 2011; **1**(3): 177-180
[9] Kojan S, Alothman A, Althani Z, Alshehri A, Mansour N, Khathaami A, et al. Granulomatous myositis associated with brucellosis; a case report and literature review. *Muscle & Nerve* 2012; **2**.DOI: 10.1002/mus.22261.