Case Report

Bilateral sacroiliitis: a rare cause for a common presentation

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ABSTRACT

Fever and osteoarticular involvement of axial skeleton are hallmark clinical manifestations of brucellosis but other diseases like ankylosing spondylitis, tuberculosis, Paget’s disease of bone, sarcoidosis should also be considered as differential diagnosis. Brucellosis is an uncommon infectious disease in India. Signs and symptoms of the disease are highly variable, with musculoskeletal complaints occurring frequently. Authors hereby reported a case of brucellosis presenting with fever, polyarthralgia, bilateral sacroiliitis and enthesitis. Knowledge of geographical distribution, evidence of exposure to the cattle and a high degree of clinical suspicion of brucellosis are very important for early differentiation from other diseases to prevent lethal complications and institute early treatment and a prompt recovery.

Keywords: Brucellosis, Bilateral sacroiliitis, Enthesitis, Fever

INTRODUCTION

Brucellosis is a zoonotic infection caused by Brucella, small gram-negative coccobacilli.\(^1\) In brucellosis, arthralgia, myalgias, and back pain often occur with the fever, which is acute, chronic or relapsing.\(^2\) Sacroiliac infection is extremely rare, only accounting for 1-4% of bone and joint infections.\(^3\) This is generally unilateral but rarely bilateral infections have been reported.\(^4\) In a recent study, polyarticular involvement have been reported in 17% of the patients with brucellosis; with sacroiliitis and peripheral arthritis in 4 patients, sacroiliitis and spondylodiscitis in 10 patients, and sacroiliitis and bursitis in one patient.\(^5\) As far as we know, our patient is the first case in the literature where a sacroiliitis. And enthesitis are simultaneously observed.

CASE REPORT

A 32-year young male, photographer by profession, resident of rural area complained of high grade, intermittent pattern fever for 3 month. Fever persisted despite being prescribed multiple oral antibiotics. Patient also had low backache for a period of approximately 2 months and multiple joints pain for 1 month which involved both large joints as well as small joints. The pain was worse while walking. Patient also reported of an ache and limitation of movement in the back and hip area. Despite of analgesics, there was no relief from pain.

There was no history of genital ulceration, swelling or increased heat in the joints. There was no history of drug abuse and promiscuous behaviour.

On examination, his core temperature was 103°F. Patient pulse rate was 94/min, regular and blood pressure was 120/80mmHg. Locomotor examination showed tenderness over multiple joint (spine, wrist, metacarpophalangeal, hip, knee, ankle, metatarsal joint). Swelling of bilateral knee joints was noted, although no obvious deformity of any joint was there. Rest of general and systemic examination revealed no obvious
abnormality. Fundus examination was within normal limits.

Laboratory tests revealed 11.5 gm% hemoglobin and total leucocyte count of 11,600/mm³ with normal differential counts and platelets. The erythrocyte sedimentation rate was 32mm/1st hour.

Liver function, urine routine and microscopy, renal function tests were all within normal limits. Infectious serology and antigen testing including dengue, malaria, chikungunya, salmonella, scrub typhus, HBsAg, anti HEV, anti HCV and HIV were negative. Blood and urine culture were sterile. Montoux test was negative. C-reactive protein was raised (35mg/dl) while rheumatoid factor, Anti-nuclear Antibodies (ANA) and HLA-B27 were negative. Bone marrow aspiration showed normal study. X-ray of chest was normal while x-ray of pelvis showed right sided sacroiliitis (Figure 1).

Figure 1: X-ray of pelvis showed right sided sacroiliitis.

Contrast-Enhanced Computed Tomography (CECT) of abdomen and thorax were normal while Magnetic Resonance Imaging (MRI) of spine and pelvis showed bilateral sacroiliitis with hip and pelvic enthesitis (Figure 2). Interestingly, brucella serology (IgM and IgG both) turned out to be positive.

Reactive brucella with sacroiliitis and enthesitis diagnosis was made and treatment was started with a combination of doxycycline (200mg/d), rifampicin (600mg/g), streptomycin (1gm/d) and indomethacin (100mg/d).

While the streptomycin treatment was ended on the 21st day, doxycycline 200mg/d, rifampicin 600mg/d and
indomethacin 100mg/d were continued until the end of the 6th week. The patient was discharged after 3 week of hospital stay.

**DISCUSSION**

*Brucella* species particularly *B. melitensis*, *B. abortus*, and *B. suis* represents a significant public health concern. Brucellosis is transmitted from animals to humans by the ingestion of infected food products, direct contact with an infected animal including cattle, goats or inhalation of aerosols. MRI is the imaging modality of choice with fat-suppressed T2-weighted and fluid-sensitive sequences for musculoskeletal brucellosis. MRI demonstrates intraarticular fluid, bone marrow edema, and periarticular involvement, especially during the early phase of the disease. Later on, subchondral sclerosis, erosions and ankylosis can occur in chronic infection setting.

In ankylosing spondylitis, there is symmetric and bilateral sacroiliitis. Unilateral changes are more common with other spondyloarthopathies. Primarily inferior (synovial) part of the joint is involved in ankylosing spondylitis. *Brucella* spondylitis, being relatively uncommon, because of fever, high ESR and osteomyelitis of the vertebral bodies the disease is often misdiagnosed and mistreated as tuberculous spondylitis in India.

Diagnostic methods for brucellosis are primarily based on serology, with the LPS smooth chains producing the greatest immunological responses in various hosts. Blood culture is the gold standard in the diagnosis of bacterial infections including brucellosis, but this method is successful in only 40-70% of the cases. The Biphasic Ruiz-Castaneda system is the traditional method for the isolation of *Brucella*. For uncomplicated brucellosis without focal involvement a 6-week course of streptomycin and doxycycline is sufficient. In cases of osteoarticular involvement, recent trials suggest a 6-month course of rifampicin, doxycycline, and streptomycin to reduce the elevated incidence of relapse.

**CONCLUSION**

The clinical manifestations of brucellosis among human beings are inconsistent and only if a high index of suspicion is maintained, will the disease be identified. Brucellosis clinically mimics other disease like tuberculosis, ankylosing spondylitis, Paget’s disease of bone, and sarcoidosis. Brucellosis is a rare cause with similar manifestation and should be kept as a differential diagnosis. Early diagnosis and prompt treatment only can prevent lethal complications.

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