Brucellosis as a Cause of Unilateral Sacroiliitis in a Haemodialysis Patient

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ABSTRACT

Brucellosis is a zoonotic disease with multiorgan involvement. Though endemic in South Asia including Pakistan, its diagnosis is often delayed because of unawareness. Herein, a case is presented of a male patient who was previously admitted and treated for culture-negative infective endocarditis in a tertiary care hospital. He later presented with Brucella sacroiliitis that responded to triple antibiotics. This case not only highlights Brucella as a cause of sacroiliitis but also points towards the importance of proper evaluation of culture-negative infective endocarditis, as retrospectively the culture-negative endocarditis in this patient was most probably caused by Brucella.

Key Words: *Sacroiliac joint, Sacroiliitis, Brucellosis, Infective endocarditis, End-stage renal disease.*

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INTRODUCTION

Brucellosis is a multiorgan zoonotic disease that can cause multiorgan granulomatous inflammation.[1] Pakistan, a South Asian country, is labelled as "possibly endemic" for brucellosis, as the disease is present in animals.[2] Unpasteurised dairy products are a common mode of transmission of this infection among travellers to areas of endemicity. Raw camel and goat milk, raw sheep or goat liver, and reindeer bone marrow have all been associated with transmission.[3] Following haematological dissemination, it can involve one or more organs. Osteoarticular involvement is one of the most common manifestations of this zoonosis.[4] Both axial and peripheral joints can be involved in brucellosis, and it is generally agreed that one of the most prevalent sites of skeletal involvement in adults is the sacroiliac (SI) joint.[5] Herein, we report a case of Brucella sacroiliitis in a patient with end-stage renal disease (ESRD), on haemodialysis, who presented after four months of being discharged from a coronary care unit for culture-negative infective endocarditis.

CASE REPORT

A 63-year male patient, diagnosed case of hypertension (HTN), and ESRD, on haemodialysis for one year, presented in the rheumatology outpatient department (OPD) with a complaint of lower backache for the last one month followed by pain and swelling in his left wrist joint for almost the same duration.

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On inquiring, he gave a history of high-grade fever for 7 days without any history of respiratory, gastrointestinal, or central nervous system complaints. Back pain was aggravated by lying supine and was minimally relieved by taking paracetamol. In past medical history, the patient was admitted in a cardiology ward for infective endocarditis about 2 months back. Three blood cultures taken at that time were negative, and he was given linezolid and cefoperazone-sulbactam for culture-negative infective endocarditis for 6 weeks. He was discharged after stabilisation when his repeat echocardiography showed healed vegetations.

Examination of the patient revealed tenderness over the left SI joint with a positive FABER (Flexion Abduction External Rotation) test. A provisional diagnosis of posterior inferior iliac spine bursitis was made but because of fever, infective sacroiliitis was kept as a differential diagnosis and the patient was admitted for further workup and treatment. Significant investigations included C-Reactive Protein (CRP) of 200 mg/l and total leucocyte count (TLC) of 31.810/cmm with 64% neutrophils. Ultrasound abdomen showed bilateral renal parenchymal disease with no visceromegaly. 2-D echocardiography showed healed vegetations on the aortic valve cusp with no evidence of new lesions/relapse of endocarditis.

X-ray of pelvis showed obliteration of the left SI joint. MRI of pelvis showed significant periarticular subchondral oedema with cystic changes and erosions at the left SI joint with surrounding periarticular soft tissue oedema and reaction extending into left anterior presacral space (Figure 1). The right SI joint was normal. The unilaterality of findings strongly suggested infectious sacroiliitis.

He was started on piperacillin-tazobactam. Since brucellosis is the most common cause of infectious sacroiliitis, the workup for brucellosis was done. Brucella antibodies were positive at titers of >1:320. Fresh blood culture was again negative. A biopsy of the left SI joint showed necrotising granulomatous inflamma-

tion. Tuberculosis (TB) Gold test was negative. Thus, the diagnosis of Brucella sacroiliitis was confirmed, and the patient was treated with renal-adjusted streptomycin, doxycycline, and rifampicin.

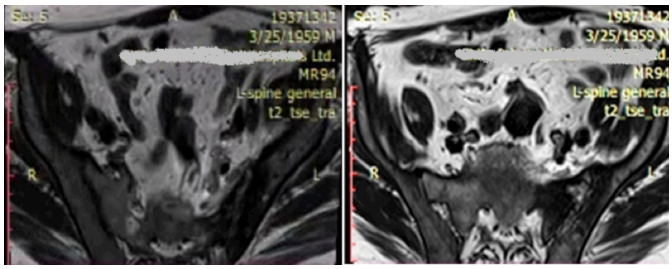

Figure 1: MRI images showing cystic and erosive changes in the left-sided sacroiliac joint.

Since the patient had a recent episode of culture-negative infective endocarditis involving the aortic valve about 2 months back, there was a high likelihood that brucellosis was the cause of that endocarditis as well.

Patient became afebrile after 1 week of treatment. Repeat titers were negative (<1:80). After 3 weeks of treatment, the patient developed cardiogenic pulmonary oedema for which he was referred to cardiology. His coronary angiography was unremarkable. Echocardiography showed healed vegetation with global left ventricle hypokinesia. He was treated conservatively with diuretics and unfortunately, all his antibiotics were stopped. Thereafter, he was lost to follow-up. However, he again reported in the OPD 2 months later with a recurrence of fever and lower back pain. Rest of the systemic inquiry was unremarkable. His CRP was again raised and Brucella abortus antibodies were again positive (1:160). Blood cultures reported after 7 days of incubation were negative. Unfortunately, the laboratory was not able to do a culture for Brucella. Based on the above history, diagnosis of relapse of Brucella sacroiliitis was made. Patient was started again on amikacin (renal dose), rifapentine (once weekly) and doxycycline (100 mg twice daily). His fever and back pain settled again with this treatment. After 4 weeks of treatment, amikacin was stopped, and rifapentine and doxycycline were continued (to complete 12 weeks). So far, the patient had no recurrence of fever or back pain or treatment-related complications.

The patient was found to be positive for both Rheumatoid factor (RAF) and anti-CCP (anti-cyclic citrullinated protein) antibodies. These tests were performed by some general practitioners that the patient went to before coming to our facility. The literature review showed that positivity for both these antibodies can be seen in patients with active Brucella infection.[6] With treatment, these antibodies become negative.

DISCUSSION

Brucellosis is a multi-organ infectious disease that can present with a wide range of symptoms. Not many cases of Brucella endocarditis were reported in the recent literature. Therefore, physicians usually do not consider this potentially lethal presentation of human brucellosis. Because brucella endocarditis is an under-recognized entity, its diagnosis is often delayed, and patients usually suffer from symptoms for 6-12 monthsbefore a diagnosis is made.[7] As with this case, the aortic valve is the predominantly affected (82%) valve in literature.[8]

Sacroiliitis due to brucellosis is another important manifestation of brucellosis. It has two-fold importance. Not only is it one of the important differential diagnoses of infective sacroiliitis but also the important differential of a much more prevalent granulomatous disease, i.e. TB, especially in Pakistan. SI joint TB accounts for approximately 5–10% of all cases of skeletal TB and thus, should be considered in TB- endemic areas like Pakistan. Besides SI joint, both brucellosis and TB can involve vertebral bodies and intervertebral disks, spondylodiskitis. In fact, Brucella spondylitis (BS) and TB spondylitis (TS) are the two leading granulomatous spinal infections.[9] In relevant age groups, infectious sacroiliitis should be differentiated from spondyloarthritis.

Treatment options include medical treatment either alone or in combination with surgical drainage for larger collections. The optimal antibiotic regimen and duration of treatment for spinal brucellosis are still controversial in the literature. The World Health Organization (WHO), in 1986, proposed antibiotic regimen of oral doxycycline, 100 mg twice a day for 6 weeks, plus oral rifampicin, 600 to 900 mg daily for 6 weeks, or streptomycin, 1 g intramuscularly daily for 2-3 weeks, for the treatment of brucellosis.[10] Even today, for uncomplicated brucellosis, the same WHO regimen is accepted as the preferred treatment by most infectious disease specialists. However, complicated brucellosis with spondylitis, endocarditis, and neurobrucellosis, requires an extended duration of treatment.

In conclusion, the infectious cause of sacroiliitis should always be considered in cases of unilateral joint involvement, and brucellosis along with TB is an important diagnosis to rule out.

PATIENT'S CONSENT:

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COMPETING INTEREST:

The authors declared no competing interest.

AUTHORS' CONTRIBUTION:

SS: Acquisition, analysis and interpretation of data.
TK: Conception of study, drafting and proofreading.
JI: Literature review and proofreading.
BA: Referencing and literature review.
All authors approved the final version of the manuscript to be published.

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