

Case Report

A case of *Brucella* sacroiliitis with abscess formation

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Introduction

Brucellosis, a multisystem infectious disease with high morbidity and low mortality, was first described by Marstan in 1860. [1]. Worldwide, approximately 500,000 cases are reported annually [2]. *Brucella* is caused by an intracellular Gram-negative bacterium [3]. Four *Brucella* species can cause human disease, *B. melitensis*, *B. abortus*, *B. suis* and *B. canis*. Human infection due to *B. melitensis* and *B. suis* is more severe. All age groups and both sexes are affected [2].

The most common mechanisms for transmission of brucellosis to humans are consumption of infected, unpasteurized animal products (raw milk, soft cheese, butter and ice-cream), skin or mucous membrane contamination with infected animal tissue or infected animal fluids and inhalation of infected aerosolized particles [2].

Brucellosis typically presents with insidious onset of fever, malaise, night sweats and arthralgia. Complications of brucellosis include infection involving one or more focal sites. Osteo-articular disease is the most common form of focal brucellosis [4]. There are several case reports published on sacroiliitis with brucellosis. Nevertheless, we report a case of a 23-year-old girl with *Brucella* sacroiliitis complicated by abscess formation who had a history of regular cheese consumption.

Case History

A 23-year-old, previously healthy girl presented with fever and low back pain for 2 weeks. She had high grade fever with chills and rigors without diurnal variation. She denied any respiratory symptoms and her bowel habits were normal. She also had difficulty in walking due to pain in her right buttock and had been unable to attend to her activities of daily living without assistance for the last 5 days. She had no other joint pains or myalgia and denied any high-risk sexual behavior. She recalled regular consumption of one kind of cheese daily for a few months. Her fever and back pain did not respond to antipyretics or simple analgesics. She was admitted to a local hospital one week after the onset of her symptoms and was treated as a case of sepsis with IV antibiotics for 5 days without any microbiological guidance. Despite treatment with conventional antibiotics her clinical condition deteriorated, and she was transferred to a tertiary care centre for further assessment.

On examination, she was febrile and ill-looking. She had moderate tenderness on the right-side sacroiliac joint with reduced range of all movements in the ipsilateral lower limb, due to pain. Her straight leg raising test was only 30 degrees without pain. She had normal motor strength, normal reflexes and intact sensation in the lower limbs. No other abnormalities were detected on nervous system examination. Cardiovascular examination showed a pulse rate of 96bpm and blood pressure of 110/80mmHg. The respiratory system and abdominal examinations were unremarkable.

Her complete blood count showed a WBC count of 11800/ μ L (Neutrophils 8800/Lymphocytes1900), Hb of 10.1g/dL with a HCT of30.4% and a platelet count of PLT 298000/ μ L. Her ESR was 128mm in the 1st hour and her CRP was80mg/dL. Her renal and liver functions were within the normal range (S. Cr-85mmol/L, Na-137mmol/L, K-4.7 mmol/L, S. Ca-2.34 mmol/L, AST-55, ALT-29, T.BIL-15 mmol/L, D.BIL-5 mmol/L, albumin-38g/L, T. protein-64g/L). As the patient was having a high ESR and CRP, sepsis was considered likely and multiple cultures were taken. However, all culture results were negative, probably due to prior treatment with antibiotics. A 2D echocardiogram was performed to exclude endocarditis and it was normal.

As the clinical examination revealed right side sacroiliac joint tenderness, a plain X-ray of bilateral sacroiliac joints was performed but no clinically significant abnormality was detected. Because of the high clinical suspicion, an MRI scan of the lumbosacral spine was arranged. Findings were diffuse T2 and STIR high signal intensities in the right side iliac and sacral articular surfaces with diffuse gadolinium enhancement. Further, it revealed erosion of both sacral articular surfaces with joint space widening. There was a 1.4×1.3cm irregular area with T1 and T2 heterogeneous signals with no enhancement after gadolinium contrast in relation to the anterior superior part of the right sacroiliac joint compatible with abscess formation (Figure 1).

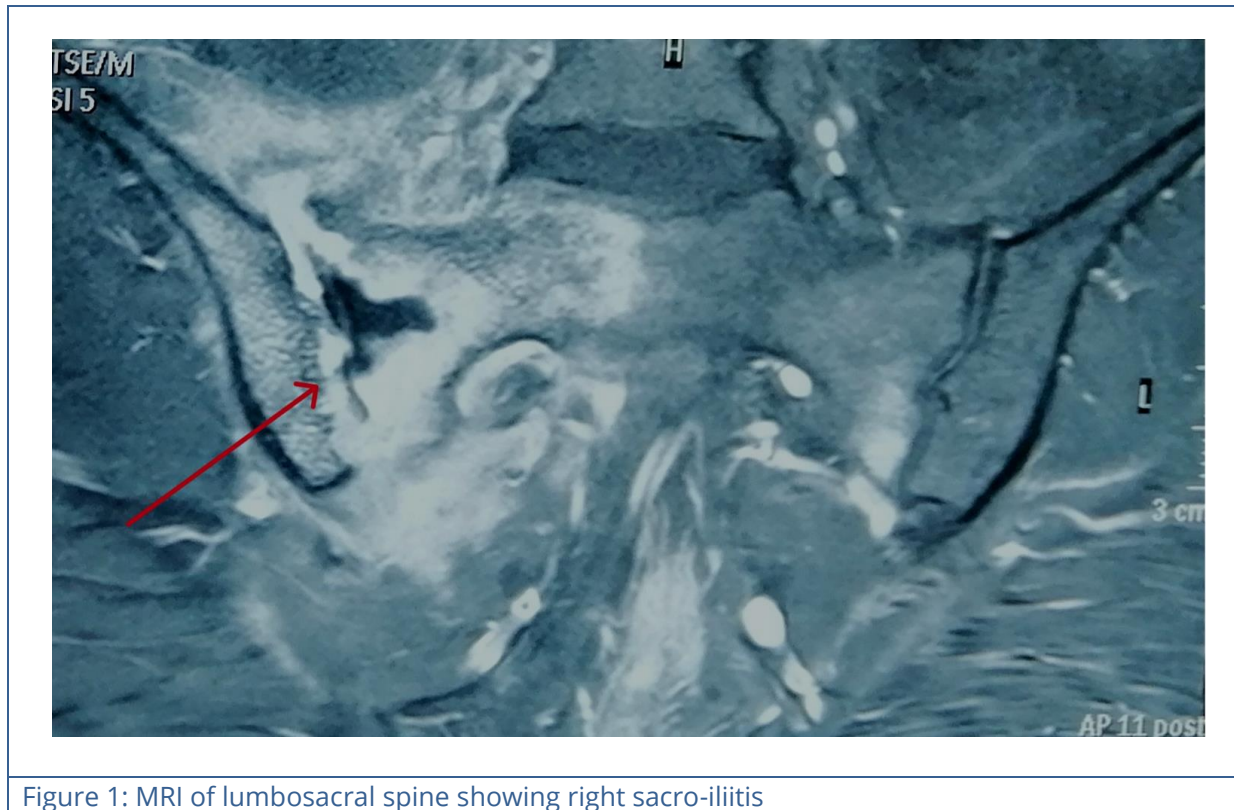


Figure 1: MRI of lumbosacral spine showing right sacro-iliitis

So, the findings were concluded as severe right side sacroiliitis with abscess formation. Melioidosis antibodies and HIV serology were negative. As there was a history of regular dairy product ingestion, a Brucella serology test was arranged which came as positive for antibodies to *Brucella melitensis*-1:320 and *Brucella abortus*- 1:320.

Prior to definitive diagnosis, she was treated with intravenous vancomycin 1g 12 hourly, intravenous meropenem 1g 8 hourly and oral doxycycline 100mg 12 hourly for 5 days and a significant clinical response was achieved with the treatment. She was discharged on oral doxycycline 100mg 12 hourly. With the availability of serological confirmation of Brucella, she was started on oral rifampicin 300mg 8 hourly and oral doxycycline 100mg 12 hourly for 6 weeks. While she was on antibiotics, USS guided drainage of the abscess in the sacroiliac joint was performed but culture did not reveal any growth. After 6 weeks, the patient was asymptomatic and her inflammatory markers became normal, hence the treatment was discontinued.

Discussion

There is hardly any literature on human infection with brucellosis in Sri Lanka. However, infection with *Brucella abortus* seems to be a major cause of abortion among cattle and buffaloes causing a major economic impact on the livestock industry. The incidence of this

disease is more prominent among animals in the dry zone. Since it is an endemic disease in Sri Lanka, there is a risk of human infection, especially among full time handlers of livestock and farm owners, as well as among consumers of infected animal products [5].

Brucellae are small, non-motile, facultative intracellular aerobic rods [2]. Out of seven zoonotic species of Brucella, four species give rise to human infection. *B. melitensis* (isolated from sheep, goats and camels), *B. abortus* (isolated from cattle), *B. suis* (isolated from swine), *B. canis* (isolated from dogs) of which infection with *B. melitensis* is the commonest [2]. Brucella is usually acquired by direct exposure to infected animal reservoirs or via ingestion of unpasteurized dairy products prepared from an infected animal, which was the probable scenario in our case [6]. Human infection can result from ingestion of as few as 10 microbes and transmission via inhalation and inoculation has also been reported. Human to human transmission is also reported rarely, by blood transfusion, tissue transplantation, breastfeeding, sexual contact, congenital transmission and nosocomial infection [2]. These pathogens affect the immune system by protein inhibition of macrophage apoptosis and by secretion of soluble tumor necrosis factor- α inhibitor [7].

The incubation period is usually two to four weeks but, occasionally, it may be long as several months [2]. Brucellosis typically presents with insidious onset of fever, night sweats (associated with a strong, mouldy, peculiar odour), malaise and arthralgia. On physical examination, hepato-splenomegaly and lymphadenopathy can be found, but these are highly variable and nonspecific.

While any organ system can be affected by brucellosis, osteoarticular disease is the commonest and accounts for 70% of patients with brucellosis [6]. In those with osteoarticular disease, the sacroiliac and spinal joints are affected commonly, 80% and 54% respectively, [2]. The knee, hip and ankle joints are sometimes affected, as well as prosthetic joints. Compared to spondylitis, both sacroiliitis and peripheral arthritis are nondestructive and heal without sequelae [6]. The exact pathophysiology of Brucella arthritis is unknown and two mechanisms have been suggested, septic and reactive forms. The septic form occurs through the blood stream as evident by isolation of brucellae from synovial fluid or tissue cultures. Brucella bursitis might be due to the reactive form. Brucella related abscess formation has also been reported, usually within the psoas or paravertebral muscles [3]. In our case, we have reported brucellosis in the sacroiliac joint complicated with abscess formation.

The second most involved organ system is the genitourinary system [7]. Neurological, cardiovascular, pulmonary, ocular and dermatological involvement with brucellosis is also reported [8]. Haematological involvement, such as mild anemia and leukopenia, can also be seen but thrombocytopaenia is far less common [1].

The diagnosis of brucellosis can be made from the typical history with epidemiological exposure plus culture of the organism from blood, body fluid or tissue or a fourfold or greater rise in *Brucella* antibody titer between acute and convalescent phase with serum specimens obtained 2 weeks apart [2]. However, the sensitivity of blood culture ranges from 53% to 90% and requires prolonged incubation. If the infection is confined to a focal area, blood culture may become negative, and serology remains the mainstay of diagnosis [6]. Literature states that a presumptive diagnosis of brucellosis can be made with either a *Brucella* antibody titer of 1:160 or more by the standard tube agglutination test of serum obtained after onset of symptoms or by detection of *Brucella* DNA in a clinical specimen by the polymerase chain reaction assay [2]. In our case, the patient presented with a typical history and exposure with focal site involvement and positive serology. Synovial fluid culture may be positive even though the blood cultures are negative [6], nevertheless our patient's synovial fluid culture was negative probably due to the delay of aspiration for over 1 month after commencement of definitive antibiotic treatment.

Radiological diagnosis of brucella focal infection is based on MRI findings. Compared to X-ray, CT and MRI have advantages like higher sensitivity, less invasive, less radiation exposure and better definition of the involvement of paravertebral and intervertebral discs [8]. In this case, X-ray of the sacroiliac joint failed to show any obvious abnormality and MRI showed better information regarding joint involvement. Another reason for this could be because the X-ray was done early in the course of illness while the MRI was performed almost 3 weeks later, and at that time infection was organized to form an abscess.

There are several antibiotic regimes for human brucellosis. A combination of doxycycline and rifampicin for 6 weeks, doxycycline for 6 weeks plus 21 days of streptomycin, doxycycline for 6 weeks plus Gentamycin for 7 to 10 days are recommended for non-spondylitis cases [9]. Doxycycline plus streptomycin combination is better out of all the regimes but feasibility may depend on several factors like the need for daily IM injection, cost and availability. Quinolone plus rifampicin also a standard regime for brucellosis and has better tolerability than doxycycline plus rifampicin [10]. Monotherapy for brucellosis is associated with a high incidence of clinical relapses and is currently not recommended [1]. *Brucella* spondylitis has a high rate of relapse and failure compared to other osteoarticular manifestations and most authorities suggest a longer duration of doxycycline therapy of at least 12 weeks [4]. In our case, we treated with a combination of doxycycline and rifampicin for 6 weeks with excellent clinical recovery without any relapses.

Conclusion

As brucellosis is an endemic animal disease in Sri Lanka there is a high risk of human disease as well. Since it has nonspecific clinical manifestations, clinicians should always consider it in their differential diagnosis if there is a strong history of exposure. Further, its osteoarticular manifestations can mimic other types of arthritis. In suspected cases blood, synovial cultures and serology tests for brucella should be considered. A prolonged course of standard

treatment must be given to prevent treatment failure or relapses. Currently, there is no available vaccine for brucella infection prevention, hence treatment of dairy products, standard precautions to minimize occupational exposure, and control of the disease in animals are the mainstay of prevention methods.

References

1. Andriopoulos P, Tsironi M, Deftereos S, Aessopos A, Assimakopoulos G. Acute brucellosis: presentation, diagnosis, and treatment of 144 cases. *International Journal of Infectious Diseases* 2007;11:52-57.
<https://doi.org/10.1016/j.ijid.2005.10.011>
2. UpToDate [Internet]. Uptodate.com2021 [cited 2021 Sep 5];Available from:
<https://www.uptodate.com/contents/brucellosis-clinical-manifestations-diagnosis-treatment-and-prevention>
3. Turan H, Serefhanoglu K, Karadeli E, Timurkaynak F, Arslan H. A case of brucellosis with abscess of the iliatus muscle, olecranon bursitis, and sacroiliitis. *International Journal of Infectious Diseases* 2009;13:e485-e487.
<https://doi.org/10.1016/j.ijid.2009.02.002>
4. Hashemi S, Keramat F, Ranjbar M, Mamani M, Farzam A, Jamal-Omidi S. Osteoarticular complications of brucellosis in Hamedan, an endemic area in the west of Iran. *International Journal of Infectious Diseases* 2007;11:496-50
<https://doi.org/10.1016/j.ijid.2007.01.008>
5. Bandara A, Mahipala M. Incidence of brucellosis in Sri Lanka: an overview. *Veterinary Microbiology* 2002;90:197-207.
[https://doi.org/10.1016/S0378-1135\(02\)00254-7](https://doi.org/10.1016/S0378-1135(02)00254-7)
6. Elzein F, Sherbeeni N. Brucella Septic Arthritis: Case Reports and Review of the Literature. *Case Reports in Infectious Diseases* 2016;2016:1-4.
<https://doi.org/10.1155/2016/4687840>
7. Priest J, Low D, Wang C, Bush T. Brucellosis and Sacroiliitis: A Common Presentation of an Uncommon Pathogen. *The Journal of the American Board of Family Medicine* 2008;21:158-161.
<https://doi.org/10.3122/jabfm.2008.02.070170>

8. Shen L, Jiang C, Jiang R, Fang W, Feng Q, Wang L et al. Diagnosis and classification in MRI of brucellar spondylitis. *Radiology of Infectious Diseases* 2017;4:102-107.
<https://doi.org/10.1016/j.jrid.2017.08.005>
9. UpToDate [Internet]. Uptodate.com2021 [cited 2021 Sep 12];Available from:
<https://www.uptodate.com/contents/brucellosis-treatment-and-prevention/print>
10. Yousefi-Nooraie R, Mortaz-Hejri S, Mehrani M, Sadeghipour P. Antibiotics for treating human brucellosis. 2021;