Skeletal lesions in a rare form of multisystemic brucellosis

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Abstract

Brucellosis is a zoonotic infection caused by bacteria belonging to the genus Brucella. Human infection typically occurs through contact with infected animals or consumption of contaminated animal products such as unpasteurized milk. In the context of rheumatic diseases, Brucellosis is known to cause musculoskeletal manifestations, making it relevant to rheumatology. The joints are commonly affected, and patients may experience symptoms such as a migratory arthritis, joint pain and myalgias. The inflammatory arthritis is often referred to as brucellar arthritis. Chronic brucellosis can be challenging in that clinical features and laboratory markers can overlap with other infectious and rheumatic diseases such as Systemic Lupus Erythematosus (SLE). This case report illustrates one of such occasions whereby chronic brucellosis presented with features mimicking a rheumatic disease and/or metastatic bone disease.

Key words*:* Brucellosis, Brucelupus, Rheumatic zoonosis, Skeletal Brucellosis, Systemic Brucellosis

Introduction

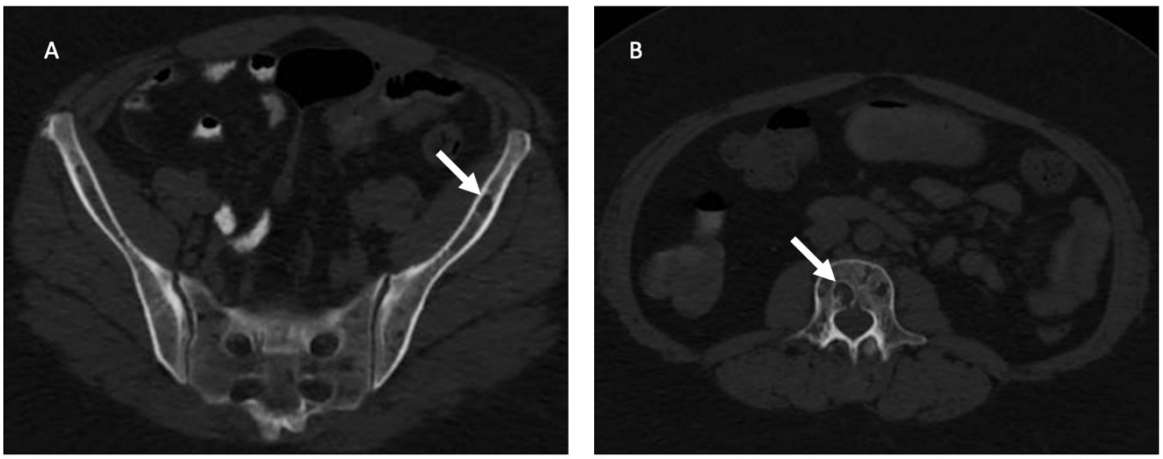
Brucellosis has been described on multiple case reports mimicking clinical features of Systemic Lupus Erythematosus (SLE), hence the coined term "Brucellupus". This case report discusses a rare presentation of the same affecting the skeletal system masquerading as either metastatic bone disease and/or autoimmune disease. Brucellosis should frequently be included in a differential diagnosis for an autoimmune-type serological and clinical presentation without an obvious diagnosis in a rural background. The medical history of the patient often is key in having a high index of suspicion for the diagnosis.

Case report

A 50-year-old unemployed female from the rural Eastern Cape Province in South Africa with a background of HIV infection on antiretroviral therapy for more than 10 years, virally suppressed, presented to the surgical department with clinical features suggestive of obstructive jaundice and degenerative spine disease. She had a 3-month duration of jaundice, coluria, recurrent axillary abscesses, and mechanical lower back pain. Her blood tests revealed an elevated ALP – 846 U/L with a normal GGT, and an elevated unconjugated bilirubin of 68 umol/L. The corrected calcium was elevated at 2.52mmol/L, haemoglobin was 10g/dL and had a normal renal function. The abdominal ultrasonography showed granulomatous lesions on the spleen and a normal liver.

Since her Antinuclear Antibodies (ANA) were found to be positive with a nucleolar pattern and a titre of 1:80, a rheumatology consult was sought. She had no clinical features to suggest a Rheumatic and Musculoskeletal Disease (RMD). The differential diagnosis upon clinical review included disseminated tuberculosis, lymphoma, multiple myeloma, metastatic disease and multisystemic brucellosis. Tests for tuberculosis done included an induced sputa for GeneXpert and a chest radiograph which were negative. A computed tomography scan of her spine and abdomen revealed multiple lytic type lesions on the thoraco-lumbar spine and the pelvic bone (Figure 1). Multiple granulomatous type lesions were observed on the spleen and mesenteric region.

Figure 1: Skeletal lesions



CT scan – bone view settings
A: White arrow demonstrating a lytic type lesion on the left pelvic bone
B: White arrow demonstrating a lytic type lesion on L5 vertebral body

A Brucella serology was done resulting in a high level of IgM and IgG for brucellosis. The serum protein electrophoresis showed a polyclonal gammopathy in keeping with a chronic infection such as HIV or chronic brucellosis. A minimally invasive mesenteric lymph node biopsy was done and showed features of a pulse granuloma. The patient was treated with Doxycycline for a month. Upon outpatient review all symptoms had disappeared and blood results for calcium levels, ALP and bilirubin had normalized.

Discussion

Brucellosis was first discovered by a team of investigators under the leadership of the surgeon Captain David Bruce in 1887 on the island of Malta. It was described to be a zoonosis capable of infecting humans through domestic animals, especially via consumption of unpasteurized milk[1]. Among RMDs, brucellosis frequently mimics SLE in terms of serological (positive ANA) and cutaneous manifestations hence the term "Brucellupus"[2].

The osteoarticular involvement can be seen in up to 85% of affected patients. The most affected sites include the sacroiliac joint (up to 80%), and the spine (up to 54%)[3]. Spondylitis and spondylodiscitis are the most frequent complication of brucella spinal involvement. Rare complications of the musculoskeletal system such peripheral arthritis, osteomyelitis, discitis, bursitis, and tenosynovitis have been reported. Spinal brucellosis has 2 forms, the focal (one vertebral level) and diffuse subtypes (multiple vertebral levels)[4,5].

The diagnosis of brucellosis remains a challenge, warranting a high index of suspicion. Ideally the diagnosis of brucellosis is made through culture of blood, bone marrow and liver biopsy. PCR is also a promising tool. The sensitivity of both culture and PCR is limited. Diagnosis is largely made through detection of specific antibodies, or by demonstrating a fourfold rise in agglutination titers in paired sera 2 weeks apart. An isolated border positive IgG may indicate past exposure to brucella or chronic brucellosis[6,7].

Case reports have highlighted the value of biopsies for making a definitive diagnosis of brucellosis, especially diagnostic splenectomies where blood cultures and PCR are negative for brucellosis[8]. An acute hepatitis secondary to brucellosis with unconjugated hyperbilirubinemia manifesting as "Gilbert-type syndrome" has been described[9], a presentation seen on the index case.

HIV infection is well known to result in positive ANA, frequently erroneously thought to be part of an RMD diagnosis[10]. Similarly, brucellosis can result in a positive ANA with some clinical features suggestive of an RMD hence the importance for clinicians to be aware of this common mimicker especially in a rural setting[11].

Conclusions

Brucellosis remains a common finding in a rural setting where domesticated animals and livestock farming is a way of life. This entity should frequently be included in a differential diagnosis for an RMD suggestive serological and clinical presentation without an obvious diagnosis. The importance of a detailed background medical history of the patient and the high index of suspicion by clinicians remain key for making the diagnosis.

Acknowledgements

We would like to thank the index patient for giving us permission to publish this case report for educational purposes.

Declarations: No conflict of interest. No funding was received to undertake the study.

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