

Unusual presentation of culture positive brucellosis

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Summary: We have reviewed the clinical presentation of 100 consecutive culture positive cases of brucellosis which came under our care during the last two years. Of these, six had atypical presentations and but for the routine practice of sending blood for brucella culture, the diagnosis would have been missed. The unusual presentations included a 19 year old boy presenting as an acute abdomen ending in laparotomy, a 52 year old man presenting with a psoas abscess, a 29 year old woman presenting with a transient perinephric mass, a 75 year old man with an acute flare up of his osteoarthritis, a 65 year old diabetic man presenting in an insulin-resistant diabetic state and a 35 year old man presenting with a cauda equina syndrome.

Introduction

Brucellosis is still one of the infectious diseases which is widespread in the Middle East, but once diagnosed is eminently treatable by appropriate antibiotics.¹ If the disease presents without well recognized features, it is possible that it may not be included in the differential diagnosis and a treatable disease will be missed. In this paper we wish to report six culture positive patients with brucellosis who presented with unusual features.

Patients and methods

We studied the clinical features of 100 consecutive brucellosis culture positive patients who were admitted to our hospital during the past two years. If the presentation did not fit in with the recognized clinical features they were grouped together as 'cases with unusual manifestation' for the purpose of this study. Amongst our 100 consecutive cases, we have come across six such patients whose case histories are reported:

Case 1

A 19 year old Saudi student presented with acute abdominal pain, fever and vomiting of two days

duration. There was diffuse abdominal rigidity and tenderness in the right iliac fossa. There was no lymphadenopathy or hepatosplenomegaly. Routine investigations showed a white cell count (WBC) of $12.3 \times 10^9/l$ with polymorphonuclear preponderance and normal biochemical profile. Acute appendicitis was provisionally diagnosed, but on laparotomy the appendix was found to be normal but there were many enlarged mesentric lymph nodes, which histologically revealed reactive lymphadenitis. Post-operatively the patient remained febrile and his blood culture had grown *Brucella melitensis* by the fifth day. The patient made a complete recovery after anti-brucella treatment.

Case 2

This 52 year old Saudi male presented with a 3 month history of low back pain, fever and night sweats. Physical examinations revealed a well built male with no pallor, clubbing or lymphadenopathy. There was a 5 cm palpable firm spleen and a palpable, non-tender cystic mass in the right iliac fossa suggestive of a psoas abscess. Routine investigations were all within normal range and computed tomographic (CT) and ultrasound scans confirmed the presence of a right-sided psoas abscess. His blood culture grew brucella and his serology for brucellosis was positive at 1/5120, thus confirming the aetiology of his psoas abscess. He responded to anti-brucella treatment with clinical and radiological disappearance of the right iliac fossa mass.

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Case 3

A 29 year old Saudi housewife presented with high fever and right sided lumbar pain without any urinary symptoms. On physical examination there was a tender palpable mass in the right lumbar region without any other positive physical findings. Routine investigations showed a WBC count of $13.5 \times 10^9/l$ with 48% polymorphs and 4% band forms and the renal profile was within normal limits. Urine microscopy showed 10 WBC/cm^3 and no red blood cells (RBC) and it was sterile. Ultrasound and CT scans confirmed the lesion to be a perinephric mass. The positive blood culture for brucella and high titre for brucella agglutination test suggested the aetiology of this perinephric mass due to brucella infection. She was put on anti-brucella treatment to which she responded clinically in a few days and there was complete clearance of the mass on follow-up ultrasound examination.

Case 4

A 75 year old Saudi man, known to be suffering from generalized osteoarthritis for the past few years, presented with severe pain and swelling of the right knee joint. He was febrile and had been immobile for one week. Physical examination showed evidence of osteoarthritis in the knees and ankles with an acutely inflamed right knee joint. Routine investigations did not show any leucocytosis and the X-rays of the knee joints showed extensive osteoarthritic changes with some areas of sclerosis. Aspiration of the right knee joint gave an exudative fluid which grew *B. melitensis* on culture. The joint fluid and blood showed high titres of brucella antibodies. The patient improved considerably and was ambulant within 10 days of commencement of anti-brucella treatment but had residual osteoarthritic changes of the joints.

Case 5

A 65 year old lean patient requiring insulin for diabetic control for the previous two years, presented in a severe uncontrolled diabetic state and weight loss. He had no symptoms suggestive of any urinary tract or chest infection and physical examination did not reveal any abnormality. He was taking combinations of human NPH and regular insulins twice daily with a total dose of more than 120 units despite which his blood sugars were in the range of 18–20 mmol/l. His urine culture, chest X-ray and haematological and biochemical investigations were all within normal limits. A bone scan showed hot spots in his right

sternoclavicular joint and one of the lower ribs. His blood culture grew *B. melitensis* and there was a rise in titre of brucella antibodies from 1/320 to 1/1280. He responded to anti-brucella treatment and his diabetic state came under control with his insulin requirements coming down to 50 units per day and a repeat bone scan showing complete resolution after treatment.

Case 6

A 35 year old Egyptian farmer, who had had a spinal injury about 10 years ago, presented with backache, weakness and wasting of the lower limbs of two months duration. Physical examination revealed no lymphadenopathy, but there was a 3 cm palpable firm splenic enlargement. Neurological examination showed wasting of both legs mainly below the knee joints with Grade IV power. Both his ankle jerks were absent with sensory loss in L5–S1 areas. Routine investigations were normal and a myelogram showed complete obstruction at the L4–L5 level due to the extradural compression. His blood cultures grew brucella organisms and in view of this spinal block he had a decompressing laminectomy. The obstructing lesion showed granulomatous tissue which was non-caseating and no acid-fast brucella could be identified. He improved completely on anti-brucella treatment and laminectomy.

Treatment

All of our patients except case 4 were treated with a combination of streptomycin and doxycycline (Vibramycin®) in doses of 1 g intramuscularly daily for 14 days and 200 mg p.o. daily for 6 weeks respectively. The duration of doxycycline was prolonged to 3 months in cases 2, 5 and 6, where there was skeletal involvement. Case 4 was not given streptomycin in view of his old age and was instead treated with co-trimoxazole (480 mg tablets – 6 tablets/day) and rifampicin 600 mg p.o. daily for a duration of 6 weeks.

Discussion

The symptoms of human brucellosis are often nonspecific, i.e. fever, chill sensations, sweating and generalized joint pains,² and the signs are usually limited to lymphadenopathy, splenomegaly and joint tenderness.^{3,4} Of the 100 culture positive cases we have studied, 94 had many of the recognised symptoms and signs enabling an easy diagnosis.

However, the six cases which we have described above were a diagnostic dilemma.

Our first case presented with acute abdominal pain mimicking appendicitis. Abdominal manifestations of brucellosis include abdominal pain, constipation, diarrhoea and hepatitis. The patient's pain was due to the acute mesenteric lymphadenitis and histology revealed hyperplasia. On reviewing the literature, a simple form of lymphadenopathy resulting from follicular hyperplasia due to acute brucellosis infections has been documented.⁵ Our case illustrates the importance of also including brucellosis in the differential diagnosis of febrile acute abdomen in an endemic area.

Our second case is a classical presentation of psoas abscess. But for the positive brucella culture and high titres of brucella antibodies, spinal tuberculosis would have been diagnosed and the patient submitted to prolonged chemotherapy. Brucellosis of the joints most often involves the lumbosacral spine, where it begins as an intervertebral disc infection and spreads to the adjacent bones, as in tuberculosis.⁶ However, the incidence of vertebral collapse and paravertebral abscess formation is lower in brucellosis than in tuberculosis.⁷ The third case presenting as a perinephric mass was a real surprise to all of us. Since we did not have a histological diagnosis of the lesion, a diagnosis of brucella could not be made with absolute certainty. However, the positive blood culture together with the clinical and radiological response to anti-brucella chemotherapy permitted us to implicate brucella as the most likely aetiological agent.

Our fourth case of painful swollen knee joints in an osteoarthritic patient is a reminder that all swollen joints in an elderly patient are not due to osteoarthritis, and the radiograph of the knee showed some areas of sclerosis, which are usually seen in skeletal brucellosis.⁸ Our fifth patient is a classical example of an ongoing infection producing uncontrollable diabetes. This patient never gave a clue regarding the possible site of infection until the bone scan showed the hot spots. Even though we did not have a tissue histology of these hot spots, the fact that they all completely disappeared on anti-brucella treatment together with the positive blood culture puts the diagnosis beyond doubt.

Our last case is an example of brucellosis producing extramedullary compression and cauda equina syndrome. Extradural compression has been attributed to a granulomatous tissue reaction extending from brucella spondylitis.⁹ This patient had a history of a spinal injury which had not produced any neurological defect but may have damaged the intervertebral disc. Once there is bacteraemia due to brucella, there is a predilection for the bacteria to settle in damaged cartilage and disks leading to the development of localized lesions.

In conclusion, we report six cases of brucellosis which presented with unusual manifestations, and but for the routine practice of brucella culture for all patients, these cases would have been missed.

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