



DIAGNOSIS AND MANAGEMENT OF AN UNCOMMON MUSCULOSKELETAL INFECTION VISWANATH

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Abstract : Melioidosis is a chronic granulomatous infection caused by a gram negative, saprophytic organism, *Burkholderia pseudomallei*. It is a cause of abscess in multiple organs and skeletal muscles. The disease is more common in susceptible individuals especially diabetics, alcoholics and immunocompromised patients. Seen more often in endemic areas, it is largely unrecognised in India. We report a series of three patients with diagnosed Melioidosis of the musculoskeletal system, stressing upon timely diagnosis, surgical and antimicrobial administration for effective treatment.

Keyword : Melioidosis, musculoskeletal, osteomyelitis, arthritis **INTRODUCTION**

Melioidosis is an infection caused by *Burkholderia pseudomallei*, a fastidious, facultative, intracellular gram negative micro-organism. Although animal species are susceptible to the micro-organism, human cases of Melioidosis are not zoonotic. Melioidosis spreads by myriad routes - direct contact with contaminated water or soil, inhalation, ingestion and cutaneous inoculation. (1) Consequently there is a multitude of clinical manifestations, giving it the moniker - 'the great imitator'. (2)

First described in 1912 by Whitmore and Krishnaswami (3), Melioidosis is an endemic disease in Southeast Asia and parts of Northern Australia. Subsequently, it has been reported in many published literature, chiefly from endemic areas of Thailand, Australia and other parts of Southeast Asia. (2,4,5)

Melioidosis causes abscesses in lung, liver, spleen, parotids and skeletal muscles, presenting as either a localised disease or as a part of a disseminated illness. (6,7) Predisposing factors for Melioidosis have been described - diabetes, alcoholism, cirrhosis and renal failure being prominent amongst them. Although, seen in other causes of an immune-compromised status, no clear association with HIV has been documented. (8)

Melioidosis of the musculoskeletal system has been studied sparingly in literature. In the musculoskeletal system, Melioidosis causes osteomyelitis, septic arthritis and intramuscular abscesses.

Reports have stressed on having a high index of suspicion especially in patients with predisposing factors, who present with a indolent osteomyelitic picture which has not responded favourably to antibiotic treatment. (5,9,10)

Most reports about diagnosis and treatment of musculoskeletal Melioidosis have been from regions of high endemicity. In India, Melioidosis was first identified in the year 1991. Subsequent reports of Melioidosis have stressed upon the fact that though India is not a region of high endemicity, Melioidosis

is a prevalent disease. However it is underreported in India due to lack of awareness, low index of suspicion and lack of sufficient facilities for its diagnosis. (11-13)

In this milieu, we seek to describe the presentation of musculoskeletal Melioidosis in three patients who presented to our department.

CASE REPORTS Case 1

Mr A, a 44 year old man was admitted for evaluation of fever with left knee joint pain, exertional dyspnea along with loss of weight and anorexia of four months' duration. On examination, he had left knee arthritis, splenomegaly, bilateral basal crepitations. The knee joint was aspirated and culture grew *Burkholderia pseudomallei*. His blood culture also grew the same organism. His bone scan showed active osteomyelitis of the metaphysis of left distal femur. He underwent decompression of left distal femur and left knee arthrotomy. (Image 1-3) Based on the sensitivity reports, he was started on intravenous Ceftriaxone which was given for 6 weeks. This was followed by oral Cotrimoxazole and Doxycycline which he continued for 20 weeks

Case 2

60 year old Mr. D presented with deep, boring pain in the region of his left thigh of one and a half months' duration. His symptoms were severe at onset and were associated with high grade fever. He was evaluated and treated by a local physician following which fever subsided, but his pain persisted. He is a known diabetic mellitus on treatment. Upon radiographic evaluation, a diagnosis of chronic osteomyelitis of the left distal femur was made. He underwent decompression and debridement of left distal femur. The material was sent for bacteriological cultures and histopathological examination.

Cultures grew *Burkholderia pseudomallei* and antibiotic sensitivity pattern was also recorded. Histopathological examination showed chronic granulomatous inflammation. (Image 4-6) He was treated with intravenous Ceftazidime for 6 weeks followed by oral Cotrimoxazole and Doxycycline which he continued for 20 weeks.

Case 3

Mr.B, a 49 year old man, presented with complaints of pain and swelling of left knee of two weeks duration. The pain was insidious onset and gradually increasing. It was initially associated with mild fever which subsided. He was treated with skin traction and immobilisation at a local hospital after which he was referred to our hospital. He had no known comorbid illnesses. Upon admission, the patient was diagnosed with septic arthritis and underwent arthrotomy of the left knee. Pus was drained and sent for bacteriological cultures, which showed presence of *Burkholderia pseudomallei*.

DISCUSSION

Melioidosis is a saprophytic infectious disease caused by *Burkholderia pseudomallei*. Melioidosis is being increasingly recognized around the world. It is far more common than previously recognized, as culture facilities for bacterial isolation are not available in most of the rural tropics, where the infection is likely to be prevalent.(8,11) Melioidosis is often associated with other underlying diseases such as diabetes, thalassemia, chronic renal failure and high alcohol consumption.(8) Two out of the three patients in our report were diabetic. Melioidosis affects all ages but peak incidence is mainly between 40 and 60 years of age, with male-to-female ratio of 1.4:1. All the three patients were belonged to this age group. Melioidosis can cause septic arthritis, osteomyelitis or soft tissue abscesses. Articular Melioidosis is relatively uncommon. It is predominantly monoarticular.(13,14) Often soft tissue abscess with osteomyelitis is associated with septic arthritis in many patients. Two of our patients had articular Melioidosis of which one was associated with osteomyelitis. Melioidosis involves the large weight-bearing joints, especially knees. There were two patients with osteomyelitis of the femur in our report. Osteoarticular Melioidosis is characterized by localized pain over underlying bone or joint, associated swelling, painful movements with fever.(15) The imaging features of osteomyelitis in long bone are soft tissue swelling, increased marrow density, osteoporosis or osteosclerosis and bone destruction; osteolytic or cystic lesions or circumscribed, saucer-shaped erosion of cortical bone with scalloped border may be evident. This mimics conditions like neoplasm, sarcoidosis and other granulomatous infections. (9,14) Any part of bone may be involved, like epiphysis, metaphysis or diaphysis; but metaphyseal region is most commonly involved. Sequestrum, periosteal new bone formation and periostitis are rarely seen. Septic arthritis and osteomyelitis show hyperemia and increased bone uptake on bone scintigraphy.

(9) Confirmation of a diagnosis of Melioidosis depends on recovering *B. pseudomallei* isolate by culture from blood, sputum, cerebrospinal fluid or other bacteriology specimen. Isolation of *B.pseudomallei* from body fluids of patients remains the 'gold standard' in diagnosis.(16,17) The main objective of treatment is to reduce mortality and morbidity and to prevent recurrent infection in Melioidosis. Melioidosis is associated with significant mortality attributable to severe sepsis and its complications. Treatment is continued over 12-20 weeks or longer if clinically indicated. This is divided into intravenous and oral phases. Ceftazidime or a carbapenem antibiotic for initial parenteral therapy should be administered for at least 10-14 days followed by a prolonged course of oral antimicrobial therapy with trimethoprim-sulfamethoxazole (TMP-SMX) with or without doxycycline. Amoxicillin-clavulanate is an alternative for children, pregnant women and for patients with intolerance to first-line therapy. Resistance of *B. pseudomallei* to these drugs is rare. A long course of oral eradication therapy is required to prevent relapse.(16) Operative intervention for musculoskeletal Melioidosis includes aspiration drainage of pus, curettage debridement, together with combination of antimicrobials. Reports

on musculoskeletal involvement of Melioidosis are few from India. A recent report on the musculoskeletal Melioidosis from a tertiary care centre emphasised on the clinical features and treatment of musculoskeletal Melioidosis in a series of five patients. (17) All three of our patients received appropriate surgical treatment along with the prescribed long term antibiotic therapy. In all patients antimicrobials were administered in two phases, the initial intravenous phase and the subsequent oral phase, for a total of 20 weeks. All patients showed clinical and microbiological recovery. Radiographs taken in the post operative visits showed healing of the osteomyelitic lesion. Laboratory parameters such as C-reactive protein levels also reduced in the follow up visit. Blood cultures taken after the completion of treatment were negative for *Burkholderia pseudomallei*.

CONCLUSION

Melioidosis is still under-recognised and under-reported in India. A high degree of clinical suspicion, prompt laboratory diagnosis, appropriate surgical management and long term bi-phasic antibiotic therapy are necessary for the comprehensive treatment of musculoskeletal Melioidosis.

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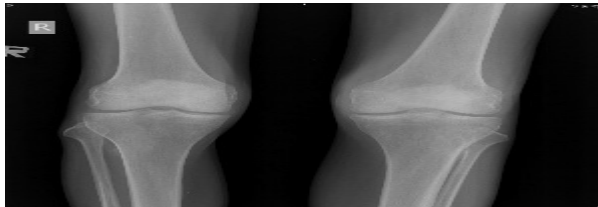


Image 1

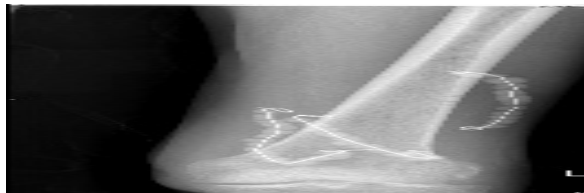


Image 2



Image 3

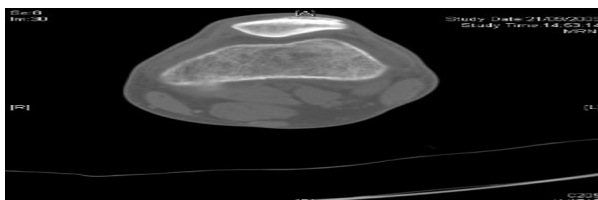


Image 4



Image 5

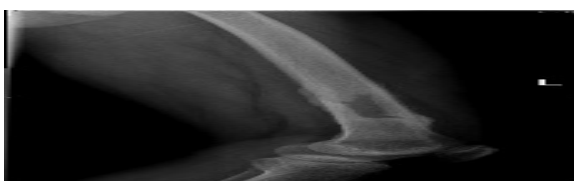


Image 6