Melioidotic septic arthritis: report of two cases

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ABSTRACT

Osteomyelitis caused by Burkholderia pseudomallei is an uncommon presentation of melioidosis. The clinical diagnosis of melioidosis is difficult as the signs and symptoms are non-specific and the duration of symptoms varies widely. We present two cases of melioidotic septic arthritis, one associated with tibial osteomyelitis and another involving the femur. Rawal Med J 2011;36:62-65).

Keyword

Melioidosis, Burkholderia pseudomallei, septic arthritis, granulomatous osteomyelitis.

INTRODUCTION

Melioidosis is an infectious disease caused by Gram-negative soil-dwelling bacillus Burkholderia pseudomallei. It is endemic in Southeast Asia and Northern Australia, with highest rates in Thailand and Australia and sporadic cases reported in other parts of Asia including India and Malaysia.¹ Mainly being a systemic disease, it rarely affects the musculoskeletal system, accounting only for 2-10% of foci of infection.²,³ Strong correlations have been postulated between melioidosis with surface water and moist clay soils, making farmers and army officers more at risk for the disease.¹
CASE 1

Mr S, a 51 years old retired army officer, a diabetic with renal impairment, presented with left knee pain for 1 year associated with intermittent fever for a month. Subsequently, knee became swollen and he was unable to walk. He was treated in a private hospital where an arthroscopic debridement was done and given a long course of antibiotics. His condition improved and was able to ambulate again after a few months of physiotherapy. He denied any contact with tuberculosis patients, exposure to muddy fields or farming.

Fig. 1a&b. Anteroposterior and Lateral view radiographs showing lytic lesions on proximal tibia; pre & post operative.
His left knee pain recurred few months later but only treated as arthritis at private clinics. He came to us with a fluctuant mass in the infrapatella area. Radiograph showed osteomyelitic changes of the proximal tibia (Fig 1a, 1b) and inflammatory markers were raised (ESR 98, CRP 18.6, WBC 17.7). Incision and drainage were done. The pus swab and tissue culture grew *Burkholderia pseudomallei*. He was immediately started on intravenous ceftazidime 1g BD. MRI showed osteomyelitic changes with communication at the proximal intercondylar notch into the knee joint (Fig 2a-c). Wound debridement, arthrotomy washout and corticotomy of the tibia were done.

**Fig 2a-c. Heterogenous lesion on proximal tibia which is hypointense on T1-weighted and hyperintense relative to muscle on T2-weighted MRI & enhanced in post gadolinium view**

The C&S of synovial fluid had no growth this time and histopathological examination revealed caseous necrosis with granuloma. Tuberculosis work up was negative and so was the bone sample sent for TB PCR. I/V ceftazidime was continued for 2 weeks followed by 20 weeks of oral trimethoprim-sulfamethoxazole and doxycycline. At
discharge, all inflammatory markers normalized and knee ROM was about 0-110°. Knee X-ray showed adequate callus formation. Outpatient clinic follow up 6 weeks later revealed he was symptom-free, with full ROM of knee and ambulating with crutches.

CASE 2

Mr B, a 40-year-old fireman with background history of diabetes, was well until he presented with prolonged fever and sepsis thus admitted to the medical ward. Thorough workup showed an incidental liver abscess and his blood culture grew *Burkholderia pseudomallei*. He was started on I/V ceftazidime and recovered from sepsis. As the abscess was small, it was treated conservatively. However, he started to complain of right knee pain which restricted his knee motion. A clinical diagnosis of septic arthritis was made. He underwent an arthrotomy washout and the tissue sample grew *Burkholderia pseudomallei* as well. At the time, he was planned for long term ceftazidime intravenously.

**Fig. 3a-c. Preoperative (a) and post operative (b) radiographs showing lytic lesions involving distal femur and latest radiograph 2 years later (c) shows severely damaged knee joint.**

Unfortunately, patient was not compliant to treatment. Though he was arranged to continue antibiotic therapy at a district hospital, there were periods of time where he
stopped the treatment. As a result, during next twelve months he had another three knee washout and debridements (Fig 3a-c). The infection then spread to the distal femur and corticotomy had to be done. Tissue culture still reveals *Burkholderia pseudomallei* infection. The infection finally resolves two years later but his knee function was badly affected.

**DISCUSSION**

Melioidosis has other synonyms such as Pseudoglanders, Vietnamese time bomb, Whitmore’s disease and Rangoon beggar’s disease.\(^1,3\) It is considered the great imitator of other infectious disease or conditions, because it can affect virtually every organ and in localized musculoskeletal infection and can mimic tuberculosis or secondary tumors.\(^4,5\) It was reported first in 1912 and thereafter it made its mark in the Vietnam War where it became a significant problem in American military medicine.\(^6\)

As the organisms are commonly found in water and soil, it is logical to correlate the infection with exposure to these factors, especially in rainy season. Preexisting medical conditions, such as diabetes mellitus, thallasemia and chronic renal failure, were often being associated with the disease in previous studies.\(^5,6\) This is attributed to the abnormal cellular immune response in diabetics, while this organism has the ability to survive in phagocytic cells.

Three modes are believed to be modes of transmission; skin being the usual portal of entry followed by inhalation and ingestion.\(^1,5,7,8\) Open wounds allow entry of this organism, which is true in one study that revealed two patients became infected when exposed to a wet rice field with an open wound on their lower limbs.\(^7\) This infection however, does not appear to be zoonosis.\(^1,7,10\) Hematogenous seeding of the organism
may result in septic arthritis, and can also spread directly from other organs or soft tissues. When it does affect a joint, it can mimic other form of infections or rheumatic disorders.

The natural course of melioidosis expands from acute, subacute or chronic, which can be days to many years, and varies from a mild pulmonary infection to overwhelming pneumonia and fulminant septicemia. Articular melioidosis is relatively uncommon and predominantly mono or polyarticular. It mainly affects large weight-bearing joints especially knees, however, shoulder joint can be affected. Osseous lesions usually involve metaphyseal regions of long bones and vertebral bodies. This is because metaphyseal region is highly vascularized, where the organism can spread hematogenously. Cystic lesion or circumscribed erosion of cortical bone can be seen on X-ray (Fig. 3). Even though MRI has a very high sensitivity and specificity for osteomyelitis, these lesions cannot be differentiated from tuberculosis and can be mistaken for other granulomatous lesions, even neoplasm.

Diagnosing melioidosis requires high index of suspicion, especially for residents and travelers of endemic areas and occupational risk factors. Isolation of the organism from body fluids through culture remains the gold standard in diagnosis. Gram staining rarely identifies the organism and histopathology might show granulomatous lesion which is indistinguishable from tuberculosis. Other methods include serological testing such as indirect hemagglutination (IHA) and enzyme linked immunosorbent assay (ELISA). Molecular method such as polymerase chain reaction (PCR) can be very useful, especially when the culture is negative.
Treatment involves long course of antibiotics that is divided into intravenous, which is the initial intensive therapy and oral phase or eradication therapy; a long term phase to prevent latency and recurrence. Choice and duration of oral antimicrobial therapy have been the most important determinants of relapse.\textsuperscript{10} Ceftazidime or carbapenem-based antibiotic is given intravenously for at least 10-14 days followed by oral trimethoprim-sulfamethoxazole (TMP-SMX) with or without doxycycline for 12-20 weeks.\textsuperscript{1,2,10} For musculoskeletal melioidosis operative intervention is needed for effective treatment. These include extensive debridement by means of drainage or curettage. Few studies recommended filling the bone defect following curettage.\textsuperscript{1,5} In conclusion, a high index of suspicion is required to diagnose melioidosis. Tuberculosis needs to be rule out when dealing with granulomatous osteomyelitis.

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\section*{REFERENCE}


