Case Report

Chronic multifocal osteomyelitis: A rare presentation of melioidosis

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Abstract

A 45-year-old gentleman presented with fever, weight loss, and painful swelling of both knees. His history was significant for type 2 diabetes mellitus. Blood cultures grew *Burkholderia pseudomallei,* and imaging revealed osteomyelitis of bilateral distal femura and proximal tibiae, with no involvement of the joint space. He underwent debridement and was initiated on ceftazidime followed by eradication therapy with trimethoprim–sulfamethoxazole. He recovered well with no further complications. Melioidosis is a rare cause of multifocal osteomyelitis and is a differential to be considered in an appropriate clinicoepidemiological setting.

Keywords: *Burkholderia pseudomallei,* melioidosis, osteomyelitis

Introduction

Melioidosis is a clinical disease that follows infection by *Burkholderia pseudomallei*.[1] Disease manifestations are protean and the organism has been described as the great mimic.[2] The spectrum of disease ranges from an acute fulminant presentation to chronic indolent disease, with diabetes being the commonest risk factor.[3] Melioidosis is endemic to Southeast Asia and Northern Australia.[4]

With a predilection to form visceral abscesses, involvement of practically every organ system has been described. Bone and joint involvement in melioidosis is a well-known entity and osteomyelitis is frequently seen in chronic disease.[5] However, multifocal osteomyelitis is rare, and available literature on the same is limited.[6,7] Chronic osteomyelitis because of *B. pseudomallei* is often misdiagnosed and treated as tuberculosis, because of the inadequacy of conventional microbiological methods.[2]

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We describe a case of multifocal osteomyelitis caused by *B. pseudomallei*. This case highlights that melioidosis is an important differential for chronic multifocal osteomyelitis, in an appropriate setting. A combined approach involving adequate surgical debridement and appropriate antibiotic therapy can yield good outcomes in chronic osteomyelitis because of melioidosis.

Case Report

A 45-year-old gentleman from North India, who worked with rail services, presented with complaints of fever for 9 months with significant loss of weight and appetite. He gave history of left knee pain and swelling, which started 3 months before presentation, followed by pain in the right knee 1 month later. His history was significant for type 2 diabetes mellitus, with good glycemic control. History was also significant for occupational exposure to soil. He underwent a debridement procedure of the left tibia and fibula in a local hospital, which revealed extensive bone destruction. He was initiated on antituberculous therapy but as his condition failed to improve, he presented to our hospital for further management.

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On examination, he was febrile, tachypneic, and tachycardic. He was pale and mildly icteric. Systemic examination was notable for hepatosplenomegaly, and musculoskeletal examination revealed swelling and tenderness over bilateral proximal tibiae with restricted knee joint mobility. Initial evaluation revealed a normal white cell count (8400 cells/mm^3, neutrophils 60%) and elevated inflammatory markers (erythrocyte sedimentation rate 65 mm, C-reactive protein 201 mg/L). Serial blood cultures grew Gram-negative bacilli, later identified as *B. pseudomallei*.

Magnetic resonance imaging of both knees revealed osteomyelitis of bilateral distal femura, proximal tibiae, and fibulae, with progressive bone destruction [Figure 1a and b] with sparing of the joint and capsule [Figure 1c]. Technetium 99-m bone scintigraphy [Figure 1d and e] showed no other sites of skeletal involvement. Imaging of the abdomen done to look for visceral abscesses revealed hepatosplenomegaly with no focal lesions.

The patient underwent a washout and debridement procedure, and per operatively, he was noted to have involvement of bilateral distal femura and tibiae, and intramuscular pus collection. Cultures taken from the necrotic tissue grew *B. pseudomallei*. Fungal and mycobacterial cultures were sterile. Histopathology of the surgical specimen was consistent with chronic pyogenic osteomyelitis. He was initiated on ceftazidime, which was continued for 6 weeks. He was also given oral trimethoprim–sulfamethoxazole eradication therapy. The fever defervesced and subsequent blood cultures remained sterile. He was progressively ambulated, and at discharge, his condition had improved. Oral eradication therapy was continued for 6 months.

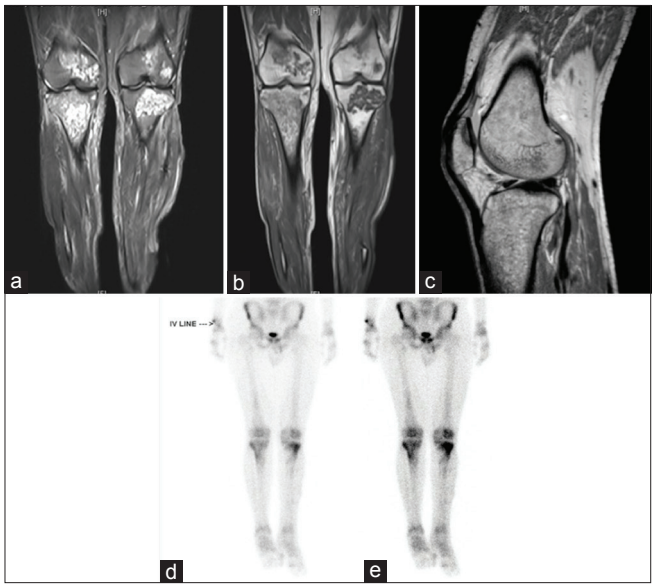

Figure 1: (a) T2W coronal image of both knees showing multifocal osteomyelitis noted as hyperintense lesions involving metadiaphyseal region of the femur and tibia bilaterally. (b) T1w coronal image showing hypointense lesions in the corresponding areas. (c) Proton density weighted MRI sequence image showing intact capsule with sparing of the joint. (d and e) triphase scintigraphy – increased tracer activity noted in both knees in the vascular, tissue and delayed phases after intravenous administration of Tc99m

He was well at completion of his eradication therapy with minimal restriction of knee joint range of movements.

Discussion

Melioidosis, caused by soil and water bacterium *B. pseudomallei*, is endemic to the tropical regions. Few instances of sporadic incidence have been reported following travel to endemic regions.[4] Recent studies have reported an increase in the cases of melioidosis diagnosed from India, owing to improved microbiological diagnostic techniques and polymerase chain reaction (PCR)-based diagnostics.[8] Identified risk factors include poorly controlled diabetes mellitus, harmful consumption of ethanol, malignancy, and chronic immunosuppression. A high clinical suspicion and adequate microbiological facilities are necessary to make a diagnosis of melioidosis.[1]

Infection is acquired by inhalation, ingestion, and inoculation, and presentation can vary from localized abscesses to frank septicemia. Chronic disease is often mistaken for tuberculosis due to endemicity of both diseases in the same tropical regions.[2] Musculoskeletal involvement in the form of osteomyelitis and septic arthritis is commonly seen, and Currie *et al.*, in a series of 540 cases of melioidosis, reported bone involvement to have an incidence of 4%.[5] Bone involvement in melioidosis can follow percutaneous inoculation or can occur in association with bacteremia, with subsequent seeding of the bone.

Multifocal osteomyelitis is a rare presentation of melioidosis. Previously reported cases have been acute in their presentation, with a mean duration of symptoms of less than 2 weeks.[6,7] This suggests that a multifocal presentation usually follows bacteremia and subsequent inoculation of multiple sites. Our patient presented with symptoms for 9 months with multifocal osteomyelitis.

Although any bone can potentially be involved in melioidotic osteomyelitis, there is a predilection for involvement of long bones, especially bones around the knee joint.[9,10] Most cases of osteomyelitis around the knee joint have associated septic arthritis. This pattern is consistent with contiguous spread of infection from one focus of seeding. Our case was unique in the fact that there was involvement of bilateral proximal tibiae and distal femura with no evidence of septic arthritis or breach of the joint capsule. This is strongly suggestive of dissemination by blood with no contiguous spread.

Cases of multifocal osteomyelitis have been noted to have poor outcomes owing to delayed diagnosis. Although common infectious etiologies such as *Staphylococcus aureus* and *Neisseria* spp. in acute settings and *Brucella* spp. and *Mycobacterium* spp. in chronic cases must be considered, the rare differential of multifocal osteomyelitis caused by *B. psuedomallei* should be kept in mind. Any delay in diagnosis and initiation of appropriate therapy could prove catastrophic. The earlier the diagnosis, the better the outcome of the patient. Hence, this needs to be

thought of at the first medical contact the patient has, which very often is the primary care physician. In settings where microbiological support is suboptimal, carefully collected blood and pus samples need to be sent to centers that have appropriate microbiological and PCR-based diagnostic assays. Once antibiotic therapy is initiated empirically, cultures become sterile and often do not yield a positive culture. These cases are often treatment-modified and misdiagnosed as tuberculosis with a higher likelihood of re-presentation (at the same site or with different organ involvement), higher incidence of septicemia, and associated mortality. It is important to identify disease early, and a combined approach with prolonged antibiotic therapy, surgical debridement, and careful monitoring can give optimal results.[11]

Our patient showed a good response to debridement and appropriate antibiotic therapy. At discharge, he was afebrile, ambulant, and advised a prolonged course of eradication therapy. This case highlights an unusual presentation of melioidosis, with multifocal osteomyelitis in the absence of septic arthritis. *B. pseudomallei* must be considered as a differential for multifocal osteomyelitis in an appropriate clinicoepidemiological setting. Primary care physicians have the best chance at making an accurate diagnosis, which weigh in significantly on the mortality and long-term morbidity associated with this infection.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

References

1. Wiersinga WJ, Currie BJ, Peacock SJ. Melioidosis. N Engl J Med 2012;367:1035-44.
2. John TJ. Melioidosis, the mimicker of maladies. Indian J Med Res 2004;119:vi-viii.
3. Cheng AC, Currie BJ, Dance DA, Funnell SG, Limmathurotsakul D, Simpson AJ, *et al.* Clinical definitions of melioidosis. Am J Trop Med Hyg 2013;88:411-3.
4. Limmathurotsakul D, Golding N, Dance DA, Messina JP, Pigott DM, Moyes CL, *et al.* Predicted global distribution of burkholderia pseudomallei and burden of melioidosis. Nat Microbiol 2016;1:15008.
5. Currie BJ, Ward L, Cheng AC. The epidemiology and clinical spectrum of melioidosis: 540 cases from the 20 year Darwin prospective study. PLoS Negl Trop Dis 2010;4:e900.
6. Nandurkar D, Lau K. Melioidosis as a cause of multifocal osteomyelitis. Clin Nucl Med 2006;31:25-7.
7. Conrad A, Valour F, Ferry T, Ader F; Lyon BJI Study Group. Multifocal melioidosis with femoral osteomyelitis in a healthy 25-year-old traveller. BMJ Case Rep 2016;2016. pii: bcr2016216356.
8. Cousins S. India is at high risk from surge in cases of melioidosis, warn researchers. BMJ 2016;352:i275.
9. Raja NS, Scarsbrook C. Burkholderia pseudomallei causing bone and joint infections: A Clinical update. Infect Dis Ther 2016;5:17-29.
10. Morse LP, Smith J, Mehta J, Ward L, Cheng AC, Currie BJ, *et al.* Osteomyelitis and septic arthritis from infection with *Burkholderia pseudomallei*: A 20-year prospective melioidosis study from northern Australia. J Orthop 2013;10:86-91.
11. Shetty RP, Mathew M, Smith J, Morse LP, Mehta JA, Currie BJ, *et al.* Management of melioidosis osteomyelitis and septic arthritis. Bone Joint J 2015;97-B: 277-82.