Case Report

Musculoskeletal melioidosis in a 33-year-old farmer presenting with the right leg cellulitis: A case report from North India

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ABSTRACT

Melioidosis, caused by *Burkholderia pseudomallei*, has been described as "the great mimicker" due to its varied clinical presentation. Musculoskeletal melioidosis is a rare presentation of this disease, and here we report a 33-year-old male, farmer from Madhya Pradesh, who presented with fever for 2 months and right leg swelling for 25 days. He was treated elsewhere as cellulitis and managed with intravenous antibiotics and fasciotomy. Magnetic resonance imaging knee and leg showed diffuse osteomyelitis of the upper tibia with similar tiny foci and sinus tract present. A diagnosis of the right knee septic arthritis with multifocal osteomyelitis was made. Focal pus culture grew pansensitive *B. pseudomallei*. He was started on Inj. Meropenem and oral cotrimoxazole tablet. He also underwent right tibial debridement, sequestrectomy, and saucerisation. He was discharged after initial therapy and has improved significantly on follow-up. This case highlights a very common clinical condition caused by a rather uncommon etiological agent and therefore renumerates the importance of insightful clinical suspicion with appropriate use of investigations and treatment options to prevent further morbidity and mortality.

Key words: Antibiotics, *Burkholderia pseudomallei*, Cellulitis, Melioidosis, Musculoskeletal infection

Melioidosis is a potentially fatal disease caused by the Gram-negative bacterium *Burkholderia pseudomallei*. It is called "the great mimicker" due to its varied clinical presentation [1]. While pneumonia and bacteremia are the most common forms, they may manifest as distant abscesses. The overall burden of this disease is grossly underestimated due to the lack of awareness and difficulty in diagnosis, especially in low-resource laboratories [2]. Early detection and appropriate antibiotic therapy can decrease morbidity and mortality to a great extent [3]. The following case highlights the clinical mimicry and difficulty in identification in the Indian context.

CASE REPORT

A 33-year-old male farmer, a resident of Gwalior, Madhya Pradesh, developed low-grade intermittent fever for the 2-month duration which began in August 2022. One month later, he developed swelling and redness in his right knee that progressed distally to involve the entire leg. There was no history of preceding trauma. Concomitantly, he was also diagnosed to have uncontrolled diabetes mellitus (glycated hemoglobin – 14.4%). He was initially evaluated at a local health-care facility where he was suspected to have right leg cellulitis. Thereafter, he underwent right leg fasciotomy and was treated with injectable Piperacillin-Tazobactum. However, he did not show significant clinical improvement and was thus referred to our facility.

On examination, he was afebrile and hemodynamically stable. His right leg was erythematous, swollen, and tender to palpation. He was not able to bear weight on his right leg. The previous fasciotomy wounds were also present along with purulent discharge. A clinical diagnosis of non-healing right leg cellulitis was made initially.

Laboratory investigations revealed mild thrombocytosis (Hemoglobin: 11.8 g/dL, WBC count: 8042 cells/cumm, and Platelet count – 6.62 lac cells/cumm) and significantly elevated C-reactive protein levels (201 mg/dL). Magnetic resonance imaging (MRI) of the right knee and leg revealed diffuse osteomyelitis of the upper tibia and sinus tract formation with the presence of similar tiny foci in the distal tibia as well (Fig. 1). A radiological diagnosis of the right knee septic arthritis with multifocal osteomyelitis was made. Pus culture done using MALDITOF-MS analyzer revealed the presence of *B. pseudomallei*, thereby confirming a diagnosis of musculoskeletal melioidosis.

The patient's antibiotics were changed to a combination of meropenem and trimethoprim/sulfamethoxazole. He also

Access this article online	
Received - 08 August 2023 Initial Review - 21 August 2023 Accepted - 12 September 2023	Quick Response code
DOI: 10.32677/ijcr.v9i10.4200	

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underwent debridement, sequestrectomy, and saucerisation of the right tibia in view of diffuse osteomyelitis. He was subsequently discharged on a 6-week course of intravenous antibiotics, followed by an oral course of trimethoprim/sulfamethoxazole and doxycycline for 6 months. Optimal glycemic control was achieved using basal-bolus insulin regimen while he was hospitalized.

The patient responded well to the treatment and eventually regained his leg function. He completed his full course of antibiotics and was followed up for 6 months, during which he remained asymptomatic. He is able to carry out his activities of daily living independently and also restarted visiting farms for work. On assessment, his wound had healed well and there were no signs of relapse. His glycemic control was adequate and he was switched to oral anti-diabetic agents. The whole timeline of the patient is depicted in Fig. 2.

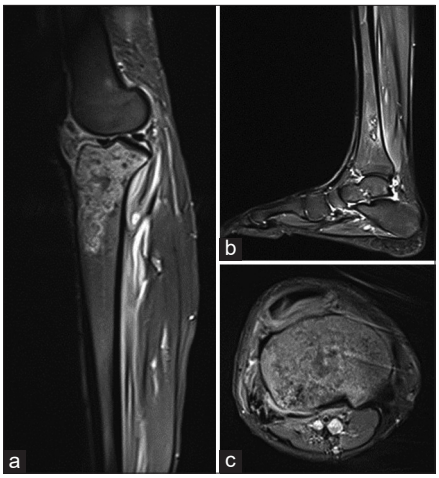

Figure 1: (a) Magnetic resonance imaging if the right leg showing septic arthritis with multifocal osteomyelitis (in clockwise manner); (b) sagittal section showing involvement of the right knee joint plus upper tibia and the lower end of tibia respectively; and (c) cross-section of the upper tibia

DISCUSSION

The above-described patient presented with a very common clinical condition caused by a rather uncommon etiological agent. Cellulitis is one of the most common surgical ailments encountered in the hospital [4]. Our approach to the diagnosis and management of this patient was based on the clinical syndrome initially; however, it was not limited to routine evaluation and empirical antibiotics since the patient had already failed treatment elsewhere. We, as any other practicing physician, also kept cellulitis as the top differential. However, since his symptoms had begun from the knee joint and then spread distally without any significant trauma, we speculated the possibility of a deeper source of infection. His occupation, recent autumn season, and immunocompromised status due to the uncontrolled diabetes mellitus predisposed him to the possibility of tropical infections such as melioidosis. This condition is hyper-endemic in South-east Asia and endemic in India [3]. However, there is a strong geographical predisposition as the majority of the cases are reported from the southern and coastal states of India [5]. Our patient belonged to the state of Madhya Pradesh, in the center of India, which is not known to harbor a huge burden of such cases. Seroprevalence and model-based studies from such areas have projected a far worse predicament than ever reported [6]. The incidence of cases has been linearly correlated with the burden of rainfall and the ever-growing epidemic of diabetes in India [7]. The gross underestimation of disease burden in various parts of India has been postulated to be due to its varied clinical presentations mimicking other etiologies, decreased awareness among doctors, and poor laboratory infrastructure, especially in the rural setting [3].

Melioidosis is considered "the great mimicker" due to its varied clinical presentation [1]. It presents most commonly as septicemia and pneumonia; however, it can also form abscesses at distinct parts of the body with the musculoskeletal system being a rare site (4–14%) [3,8]. Diagnosis of melioidosis can be challenging and subtle clues from various laboratory and

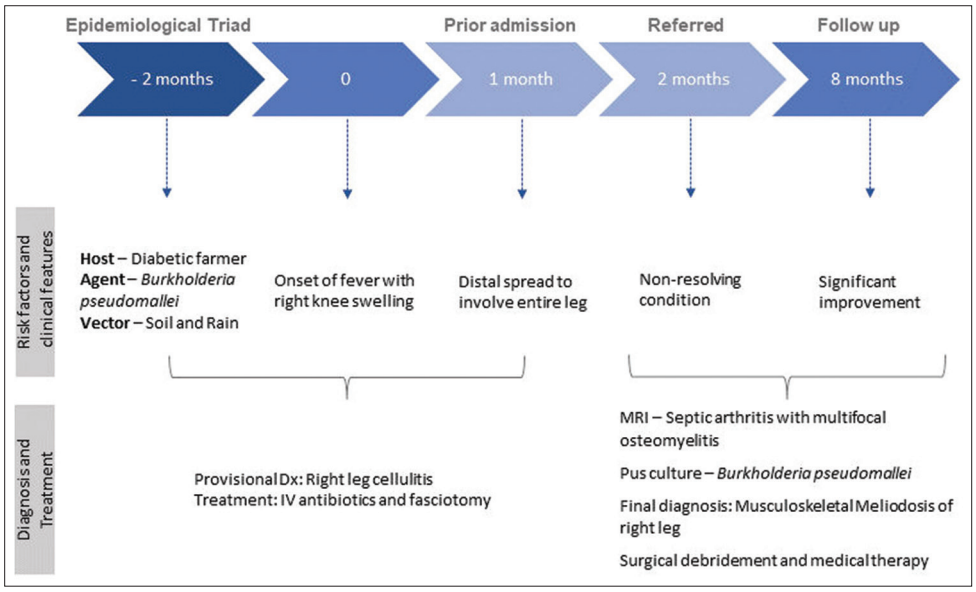

Figure 2: Timeline of the patient

radiological studies might provide valuable insights. The prompt use of higher-end radiological investigations, such as MRI in our case, provided a noteworthy clue toward his clinical conundrum. An impression of septic arthritis with multifocal osteomyelitis primarily indicates an infectious etiology but may also include other rare non-infectious etiologies such as degenerative process, vasculopathy, trauma, and even neoplastic process [9].

The role of accurate microbiological testing and assessment becomes immensely valuable. The appearance of *Burkholderai pseudomallie* on conventional culture mediums might mimic other organisms and kit-based assays often lead to misidentification [10]. We were able to correctly identify and speciate the organism due to the availability of MALDITOF-MS-based analyzers at our center [11]. In low-resource settings, antimicrobial susceptibility patterns can help to differentiate it from other Gram-negative bacteria such as pseudomonas [3]. *B. pseudomallie* possesses intrinsic resistance to various commonly used classes of antimicrobials such as penicillin, ampicillin, first and second-generation cephalosporins, and aminoglycosides and shows false positive sensitive results with quinolones. Detailed antimicrobial susceptibility testing was also done to further guide the therapy. The limitation lies in the fact that routinely the pus samples are not considered as an appropriate specimen for determining the pathogenic microorganism; however, since virulent strains of *B. pseudomallei* rarely colonizes human surface, its detection even in poor quality pus samples would warrant treatment [10].

The recommended treatment for musculoskeletal melioidosis is divided into an initial initiation phase of 6 weeks followed by the eradication phase of 6 months. During the intensive phase, intravenous (IV) ceftazidime or meropenem can be given depending on the patient's clinical condition and severity. Our patient was given IV meropenem as a limb-salvaging therapy. Oral trimethoprim/sulfamethoxazole provides better tissue penetration and is therefore added in the intensive phase as well [12]. The eradication phase mainly consists of oral trimethoprim/sulfamethoxazole therapy; however, a second agent (tablet doxycycline) was added in view of diffuse disease. Surgical therapy is indicated as a treatment option for chronic osteomyelitis. In our case, extensive surgical debridement, sequestrectomy, and saucerization of the right tibial bone provided adequate source control and thus reduced the risk of recrudescent infection.

CONCLUSION

Meliodosis should be considered as an important differential diagnosis in the setting of cellulitis and/or osteomyelitis, especially when the epidemiological triad is conducive. A strong clinical suspicion becomes even more important as the microbiological detection of *B. pseudomallei* offers a great challenge, particularly in low-resource settings. The empirical treatment for common causative organisms fails to confer any substantial improvement due to its intrinsic antimicrobial resistance. However, with correct identification, appropriate and prolonged antibiotic and surgical treatment can be offered with a substantial reduction in mortality and relapses.

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Funding: Nil; Conflicts of interest: Nil.

How to cite this article: Jindal A, Gulati S, Dessai R, Chawla K. Musculoskeletal melioidosis in a 33-year-old farmer presenting with the right leg cellulitis: A case report from North India. Indian J Case Reports. 2023;9(10):303-305.