Clinically lesser known entity in India: A Report of two cases of Melioidosis

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Melioidosis is endemic in the South Asian regions, like Thailand, Singapore Malaysia and Australia. The disease is more pronounced in the southern part of the country. It is caused by *Burkholderia pseudomallei* which causes systemic involvement, morbidity and mortality associated with the disease is high. Due to highly varied clinical presentation, and low general awareness this infection is largely underdiagnosed and under reported in our country. Most laboratories in the country still rely on conventional culturing methods with their low sensitivity, adding to the under reporting. To enhance physician awareness we describe here two cases who presented to our institute after months of misdiagnosis.

**Keywords:** *Burkholderia pseudomallei*, melioidosis, systemic involvement

**Introduction**

Melioidosis, though endemic in the Southeast Asian regions, such as Thailand, Singapore Malaysia and Australia,¹ was also reported from Africa, North and South America, Pacific and Caribbean islands, Middle east and Europe.² In India, it is more prevalent in the south; though also reported from other parts.³,⁴ The disease is underreported due to its protean manifestations and low physician index of suspicion. Many laboratories relying on conventional culture methods confuse it with *Pseudomonas* species due to common phenotypic characteristics. John et al reported that the disease could be more prevalent than what is available in literature.⁵

Melioidosis is seen more in diabetics and other immunosuppressed conditions.⁶ We report two who were undiagnosed initially and presented to us within 3 months of each other. Both belonged to Madhubani district in Bihar, raising possibilities that the area could be endemic for the disease.

**Case Report**

**Case 1**

A 65-year-old male presented with fever, cough with breathlessness for last 2 months, along with swelling, redness and pain affecting both ankles for 7 days. Two months earlier, he was treated for clinically suspected typhoid with no resolution of the fever. At the same time he was also found to be diabetic. At presentation, patient was toxic, febrile, icteric and dehydrated. Body temperature was 101 °F, BP was 90/70 mmHg, respiratory rate 50/min, heart rate 128/minute. Bronchial breath sounds were heard on the left side and bilateral crackles were present. Spleen was mildly enlarged. Both ankles were erythemic and edematous. The right foot was affected by cellulitis with purulent discharge. Investigations showed a total leukocyte count (TLC) of 24,000/mm³, the peripheral blood smear revealed toxic granulations and vacuolations, the liver enzymes were raised with the values of aspartate aminotransferase (SGOT) - 160 IU/L, alanine aminotransferase (SGPT) - 82 IU/L, alkaline phosphatase (ALP) - 1120 IU/L, G-glutamyl transferase (GGT) - 216 IU/L, lactate dehydrogenase (LDH) - 387 IU/L., A chest X-ray showed homogenous consolidation on the left side, An ultrasound showed bilateral pleural effusions and mild ascites; high sensitivity C-reactive protein (Hs CRP) level was 233.8 mg/dl, procalcitonin level was 0.5 ng/ml,
glycosylated hemoglobin (HbA1c) level was high at 11.10%, acid-fast stain (AFB) from sputum carried out for 3 days, malarial antigen, Widal test, Urine culture were all negative. Blood from two different peripheral sites and endotracheal secretions were sent for culture. Arterial blood gases showed a hypoxic state With an oxygen saturation at 78%. He was ventilated and based on a clinical diagnosis of sepsis, started on an empirical therapy with piperacillin/tazobactum and clindamycin. He continued to be febrile and on day 3, right ankle was surgically drained and pus collected for culture. Blood culture put up in the fully automated Bactec (Beckton and Dickinson) showed positivity on the third day and grew nonfermenting, oxidase-positive flat, dry, wrinkled colonies which were subsequently identified as Burkholderia pseudomallei in Microscan 96 SI (Siemens, Frimley, Camberley, UK).

Burkholderia pseudomallei was also isolated from his endotracheal secretions and pus.

Case 2

A 58-year-old male presented with fever (on and off from last 1 to 11/2 years), cough, pain and swelling of left knee for last 2 weeks. He had taken treatment from various local practitioners without success. He was a known diabetic for last few years.

On presentation, his temperature was 100 °F, pulse 90/minute; blood pressure 94/50 mmHg, respiratory rate 38/minute. Left knee was hot and tender; however, an X-ray of the knee was normal. Bilateral scattered crepitations were heard on chest examination. Oxygen saturation was 80% for which he was started on non-invasive ventilator support. A chest X-ray showed fluffy nodular opacities in both lung fields. He was empirically started on piperacillin/tazobactum.

Investigations showed – a TLC of 10,900/cubic mm, the liver enzymes were raised with values of SGOT - 117 IU/L, SGPT - 175 IU/L, ALP- 69 IU/L, GGT – 81 IU/L, LDH - 289 IU/L. HsCRP level was 166.7 mg/dl, procalcitonin level was 0.9 ng/ml. Hb A1c level was 10.0%. Sputum for AFB (stain and culture), Malaria antigen test and Widal test were all negative. Blood cultures were sent from two different peripheral sites on the day of admission. On day 3 of admission, he had spikes of fever. Another set of blood culture was sent. Burkholderia pseudomallei was isolated from all these blood samples.

Both the strains were resistant to ceftazidime (MIC>32 mg/l) and sensitive to imipenem (MIC <2 mg/l), co-trimoxazole (MIC <1/19 mg/l) and tetracycline (MIC <2 mg/l). Antibiotics were escalated to imipenem (50 mg/kg/day) in both cases. Both cases responded well and were discharged on maintenance therapy with co-trimoxazole and doxycycline for 20 weeks. Blood cultures repeated 1 month afterwards came out to be sterile. Both these strains were handled in Biosafety level-2 in laboratory.

Discussion

Melioidosis is a clinical entity ranging from acute fulminant septicemia to a chronic state. Three modes of acquisition (inhalation, ingestion, inoculation) are known. Skin and soft tissue infections may occur after minor wounds or from hematogenous spread. Immunosuppressed persons are more at risk. It is an emerging infection in India with the first case reported in a child from Dapoli in Maharashtra in 1990. It is reported also from Kerela, Karnataka and the south east. Vidyalaksmi et al. found fever to be the commonest complaint (96% cases) and diabetes mellitus as a predisposing factor which we found in both our cases. The second case had a long history of fever attributable to the chronic form of melioidosis. Both cases belonged to an area where people are exposed to flood waters. Their agricultural background could explain the exposure. Burkholderia pseudomallei is present as an environmental saprophyte in soil and fresh surface water in endemic regions posing a high risk of infection to this group of people. Lack of clinical know-how of this disease and insufficient laboratory expertise usually hamper the diagnosis of the disease. Many authors have expressed similar concern of the factors that lead to under reporting. Therefore, it is worthwhile to document these cases.

Both strains had similar antimicrobial susceptibility pattern hinting toward a common source. Further epidemiological studies are indicated to determine the geographical prevalence and risk factors of this condition.

References

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Source of Support: Nil, Conflict of Interest: None declared.