

CASE SERIES OF *BURKHOLDERIA PSEUDOMALLEI* CAUSING MELIOIDOSIS (PULMONARY, CEREBRAL, AND SPLENIC ABSCESS)PRASANNA S^{1*}, MAYURI MAHAJAN², NIKHIL MAHAJAN³

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ABSTRACT

Burkholderia pseudomallei is soil saprophytic Gram-negative bacilli that cause a fatal disease called melioidosis. Melioidosis is capable of causing cutaneous infection and systemic infections in the respiratory tract, cardiovascular, gastrointestinal, urinary, skin and soft tissue, and musculoskeletal and central nervous systems. Here, we report rare forms of pulmonary, cerebral, and splenic abscess case series of melioidosis caused by *B. pseudomallei*. Imported cases have been reported among tourists, immigrants, and soldiers who returned from endemic areas. The acquisition of infection is through percutaneous, inhalation, and ingestion of contaminated water; person-to-person transmission is very rare. Melioidosis cases are primarily found in the rainfall season and are usually associated with risk factors such as diabetes, alcoholism, and chronic renal diseases. However, 20–26% of cases were not associated with predisposing conditions. The identification is based on colony morphology, Gram stain, antibiotic susceptibility testing, and other supportive automated and molecular assays when we suspect *B. pseudomallei*. There are two phases, the intensive and eradication phases, in managing melioidosis. In the intensive phase, ceftazidime for 2 weeks showed efficacy in almost 50% of cases, and the eradication phase treatment with co-trimoxazole and doxycycline or amoxicillin/clavulanic acid for 3–6 months showed an excellent response. The improper clinical diagnosis and management of *B. pseudomallei* can lead to complications. Hence, early diagnosis with microbiological approaches such as culture, biochemical reactions, or automated systems available and antimicrobial sensitivity testing will cure the patient quickly without mortality.

Keywords: *Burkholderia pseudomallei*, Melioidosis, Respiratory, Cerebral, and Splenic abscess.

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INTRODUCTION

Burkholderia pseudomallei is soil saprophytic Gram-negative bacilli that cause a fatal disease called melioidosis. In 1912, Whitmore and Krishnaswami (Burma) first described and isolated *B. pseudomallei* [1,2]. Other countries such as Malaysia, Singapore (1913), and Vietnam (1925) also reported cases of melioidosis later. Cases of melioidosis or other forms of *B. pseudomallei* have been reported in other Southeast Asian countries and Australia. Furthermore, various case reports have been notified from Kerala, Tamil Nadu, and eastern and north-eastern parts of India [3-5].

B. pseudomallei is ubiquitous and present in soil and water and can cause abscesses in solid organs. Melioidosis is capable of causing cutaneous infection and systemic infections in the respiratory tract, cardiovascular, gastrointestinal, urinary, skin and soft tissue, and musculoskeletal and central nervous systems. The most typical clinical presentation will be affecting the respiratory and cardiovascular systems. Respiratory system complications such as pleuritic and pulmonary abscesses are common following pneumonia, and in the cardiovascular system, bacteremia, pericarditis, septic emboli, and mycotic aneurysms [2,3]. Here, we report rare forms of pulmonary, cerebral, and splenic abscess case series of melioidosis caused by *B. pseudomallei*.

CASE DESCRIPTIONS**Case 1**

A 50-year-old male presented to the emergency department with fever, abdominal pain, and referred pain over the right side of the shoulder. The patient was a known diabetic and not a known hypertensive. On clinical examination, the patient had a throbbing type of pain with warmth below the right costochondral. After further investigation, the patient was diagnosed with a splenic abscess with type 2 diabetes mellitus. He was admitted to the intensive care unit (ICU), and other

routine investigations were done. The nasopharyngeal swab was sent for RT-PCR and turned out to be coronavirus disease (COVID-19) negative. All blood investigations were within normal range except blood glucose, increased leukocytes, and raised C-reactive protein. The patient was given insulin and empirical therapy with a piperacillin and tazobactam combination. The blood and urine samples were sent for culture and sensitivity and were negative. The ultrasonogram (USG) of the abdomen done shown a splenic abscess, USG-guided splenic aspiration was performed, and pus was sent for culture and sensitivity. Gram stain of splenic aspirate showed numerous pus cells with safety-pin appearance gram-negative bacilli (Fig. 1).

Case 2

A 55-year-old male presented to the general medicine outpatient department with breathlessness and expectorating cough for 2 weeks. The patient was a known case of diabetes and was on antidiabetic drugs. On clinical examinations of the respiratory system, there was wheeze and crackles; other system examinations were within normal limits. The patient was diagnosed with acute bronchitis with secondary pneumonia. He was admitted, and a sputum sample was sent for culture, Gram stain, acid-fast stain, and other routine investigations. The nasopharyngeal swab was sent for RT-PCR and turned out to be COVID-19 negative. All blood investigations were within normal range except blood glucose. The patient was given insulin and empirical antibiotics (piperacillin-tazobactam). The sputum sample was negative for the Ziehl-Neelsen stain; the gram stain showed numerous pus cells with safety-pin appearance gram-negative bacilli and KOH mount, and Gomori's methenamine silver stain showed no fungal elements.

Case 3

A 30-year-old male presented to the emergency department with a headache and altered sensorium for a 1-week duration. The patient was not have a known case of diabetes and hypertension and had no

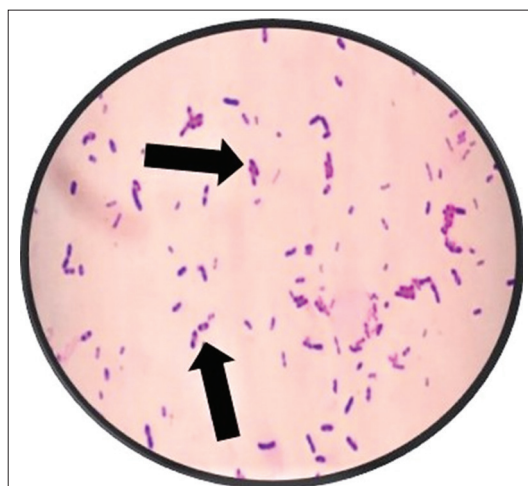


Fig. 1: Gram stain showing a safety-pin appearance of *Burkholderia pseudomallei*

history of head injury. On clinical examination of the system, he was a suspected case of meningitis and was admitted to the medical ICU. The lumbar puncture was done under aseptic precaution; a cerebrospinal fluid (CSF) sample was sent for culture, Gram stain, and acid-fast stain, and other routine investigations were done. The nasopharyngeal swab was sent for RT-PCR and turned out to be COVID-19 negative. All blood investigations were within normal range. The patient was given empirical treatment with cefotaxime 1 g IV BD. The CSF sample was negative for ZN stain; Gram stain showed pus cells with safety-pin appearance gram-negative bacilli and KOH mount, and Gomori's methenamine silver stain led no fungal elements.

Management of all cases

The sample was cultured in blood agar, MacConkey, and Chocolate agar identified as *B. pseudomallei* based on colony morphology, biochemical (Figs. 2 and 3), and VITEK-2, and was sensitive to cotrimoxazole, ceftazidime, ceftazidime-sulbactam, and meropenem. The patient was managed with a piperacillin and tazobactam combination initially and switched over to ceftazidime-sulbactam after sensitivity testing.

DISCUSSION

Melioidosis is a deadly infection caused by the ubiquitous pathogen *B. pseudomallei* seen in soil and water. In non-endemic regions, melioidosis is a fatal infection that remains unknown to the general population and health-care workers [8]. Southeast Asia and northern Australia remain to report leading cases of melioidosis, with the highest number of cases, 2000–3000 each year, said in Thailand [9]. Imported cases have been reported among tourists, immigrants, and soldiers who returned from endemic areas. The acquisition of infection is through percutaneous, inhalation, and ingestion of contaminated water; person-to-person transmission is very rare [10]. Melioidosis cases are primarily found in the rainfall season and are usually associated with risk factors such as diabetes, alcoholism, and chronic renal diseases. However, 20–26% of cases were not associated with predisposing conditions [11,12]. Some of the presentations of melioidosis are septicemia, localized infection, asymptomatic infections, ulcers, pneumonia, visceral abscesses, neurologic infection, and musculoskeletal infections [13]. The specimens such as blood, pus, wound swabs, urine, and sputum from which this organism can be isolated, and the identification up to *species* level as *B. pseudomallei* is not usually done with manual and some automated identification system. Henceforth, the diagnosis is based on colony morphology, Gram stain, antibiotic susceptibility testing, and other supportive automated and molecular assays when we suspect *B. pseudomallei*. The worldwide problem is the similarity of this organism to *Pseudomonas species* and the delay in identifying as well as initiation of treatment [14]. *B. pseudomallei* is resistant to antibiotics such as penicillin, ampicillin, first- and second-generation cephalosporins, macrolides, rifampicin, and



Fig. 2: (a) Blood agar showing irregular margined, large, and moist colonies; (b) MacConkey agar showing non-lactose to late lactose fermenting colonies; (c) wrinkled and rugose appearance in MacConkey

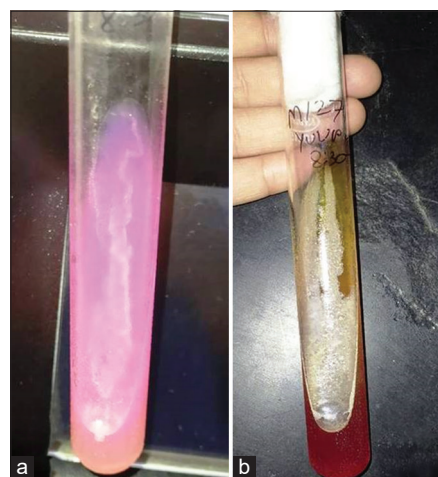


Fig. 3: (a) Urea hydrolysis and (b) Triple-sugar iron agar showing non-fermentative growth

aminoglycosides. It is sensitive to cefotaxime, ceftriaxone, ceftazidime, imipenem, meropenem, piperacillin, and amoxicillin/clavulanate in combination [15]. There are two phases, the intensive and eradication phases, in managing melioidosis. In the intensive phase, ceftazidime for 2 weeks showed efficacy in almost 50% of cases, and the eradication phase treatment with co-trimoxazole and doxycycline or amoxicillin/clavulanic acid for 3–6 months showed an excellent response [16–18].

CONCLUSION

The highlight is to know the importance of melioidosis in the Indian and Asian populations and its different manifestations. The inappropriate clinical diagnosis and management of *B. pseudomallei* can lead to complications. Hence, early diagnosis with microbiological approaches such as culture, biochemical reactions, or automated systems available and antimicrobial sensitivity testing will cure the patient quickly without mortality.

CONSENT

Consent was taken from patients for publication purposes, and personal identity is not exposed.

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Nil.

AUTHORS CONTRIBUTION

First and corresponding author contributions: Lab diagnosis, complete write-up, photos, editing, and compiling; Second author contributions:

Lab diagnosis and reporting; Third author contributions: Supportive clinical diagnosis.

CONFLICT OF INTEREST

Nil.

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REFERENCES

- Currie BJ. *Burkholderia pseudomallei*, *Burkholderia mallei*: Melioidosis and glanders. In: Mandell GL, Bennett JE, Dolin R, editors. Mandell, Douglas and Bennett's Principle and Practice of Infectious Disease. 7th ed. Philadelphia, PA: Churchill Livingstone; 2010. p. 2869-77.
- Vernon M. Pseudomonadaceae. In: Minor LE, Vernon M, editors. Bacteriologiemédicale. 2nd ed. Paris: Flammarion; 1989. p. 555-98.
- Anuradha K, Meena AK, Lakshmi V. Isolation of *Burkholderia pseudomallei* from a case of septicaemia: A case report. Indian J Med Microbiol 2003;21:129-32. doi: 10.1016/S0255-0857(21)03138-8, PMID 17642999
- Jesudason MV, Anbarasu A, John TJ. Septicaemic melioidosis in a tertiary care hospital in south India. Indian J Med Res 2003;117:119-21. PMID 14575177
- Viswaroop BS, Balaji V, Mathai E, Kekre NS. Melioidosis presenting as genitourinary infection in two men with diabetes. J Postgrad Med 2007;53:108-10. doi: 10.4103/0022-3859.32210, PMID 17495376
- Kosuwon W, Taimglang T, Sirichativapee W, Jeeravipoolvarn P. Melioidotic septic arthritis and its risk factors. J Bone Joint Surg Am 2003;85:1058-61. doi: 10.2106/00004623-200306000-00011, PMID 12784002
- Wiersinga WJ, Virk HS, Torres AG, Currie BJ, Peacock SJ, Dance DA, et al. Melioidosis. Nat Rev Dis Primers 2018;4:17107. doi: 10.1038/nrdp.2017.107, PMID 29388572
- Dance DA. Melioidosis as an emerging global problem. Acta Trop 2000;74:115-9. doi: 10.1016/S0001-706X(99)00059-5, PMID 10674638
- Vuddhakul V, Tharavichitkul P, Na-Engam N, Jitsurong S, Kunthawa B, Noimay P, et al. Epidemiology of *Burkholderia pseudomallei* in Thailand. Am J Trop Med Hyg 1999;60:458-61. doi: 10.4269/ajtmh.1999.60.458, PMID 10466977
- Inglis TJ, Garrow SC, Henderson M, Clair A, Sampson J, O'Reilly L, et al. *Burkholderia pseudomallei* traced to water treatment plant in Australia. Emerg Infect Dis 2000;6:56-9. doi: 10.3201/eid0601.000110, PMID 10653571
- Currie BJ, Jacups SP. Intensity of rainfall and severity of melioidosis, Australia. Emerg Infect Dis 2003;9:1538-42. doi: 10.3201/eid0912.020750, PMID 14720392
- Cheng AC, Currie BJ. Melioidosis: Epidemiology, pathology, and management. Clin Microbiol Rev 2005;18:383-416. doi: 10.1128/CMR.18.2.383-416.2005, PMID 15831829
- Dhodapkar R, Sujatha S, Sivasangeetha K, Prasanth G, Parija SC. *Burkholderia pseudomallei* infection in a patient with diabetes presenting with multiple splenic abscesses and abscess in the foot: A case report. Cases J 2008;1:224. doi: 10.1186/1757-1626-1-224, PMID 18840270
- Dancet DA. Melioidosis: The tip of the iceberg? Clin Microbiol Rev 1991;4:52-60.
- Ashdown LR. *In-vitro* activities of the newer β -lactam and quinolone antimicrobial agents against *Pseudomonas pseudomallei*. Antimicrob Agents Chemother 1988;32:1435-6. doi: 10.1128/AAC.32.9.1435, PMID 3196005
- White NJ, Dance DA, Chaowagul W, Wattanagoon Y, Wuthiekanun V, Pitakwatchara N. Halving of mortality of severe melioidosis by ceftazidime. Lancet 1989;2:697-701. doi: 10.1016/S0140-6736(89)90768-x, PMID 2570956
- Chetchotisakd P, Chierakul W, Chaowagul W, Anunnatsiri S, Phimda K, Mootsikapun P, et al. Trimethoprim-sulfamethoxazole versus trimethoprim-sulfamethoxazole plus doxycycline as oral eradication treatment for melioidosis (MERTH): A multicentre, double-blind, non-inferiority, randomised controlled trial. Lancet 2014;383:807-14. doi: 10.1016/S0140-6736(13)61951-0, PMID 24284287
- Inglis TJ. The treatment of melioidosis. Pharmaceuticals (Basel) 2010;3:1296-303. doi: 10.3390/ph3051296, PMID 27713302