EDORIUM Journals

Int J Case Rep Images 2019;10:101049Z01ST2019.
www.ijcasereportsandimages.com

CASE REPORT PEER REVIEWED | OPEN ACCESS

Melioidosis presented as severe acute cholangitis: A rare presentation

Syazwani Mohd Tajudin, Alwi Muhd Besari, Nabilah Ismail, Siti Asma' Hassan, Abdul Rahman Zaidah

ABSTRACT

Introduction: Melioidosis is caused by a Gram-negative bacteria known as *Burkholderia pseudomallei*. Clinical manifestation of the disease is unspecific, leading to wrong diagnosis and delayed in administration of appropriate antibiotics. Case Report: This case reported an atypical presentation of melioidosis in a 45-year-old Malay lady with underlying asthma and hypertension who presented with generalized abdominal pain for 14 days. A few days before admission she developed high grade fever and jaundice. She was initially treated as ascending cholangitis. The diagnosis was changed to disseminated melioidosis after computed tomography (CT) scan findings and isolation of *B. pseudomallei* from the blood culture. She was treated with antibiotics according to the melioidosis regime and responded well to the therapy. Conclusion: Acute cholangitis or cholecystitis is a rare presentation of melioidosis. High degree of suspicion is very crucial in a patient presented with any severe illness especially in an endemic area.

Keywords: *Burkholderia pseudomallei*, Cholangitis, Melioidosis

How to cite this article

Tajudin SM, Besari AM, Ismail N, Hassan SA, Zaidah AR. Melioidosis presented as severe acute cholangitis: A rare presentation. Int J Case Rep Images 2019;10:101049Z01ST2019.

Article ID: 101049Z01ST2019

****

doi: 10.5348/101049Z01ST2019CR

Syazwani Mohd Tajudin[1], Alwi Muhd Besari[2,3], Nabilah Ismail[3,4], Siti Asma' Hassan[3,4], Abdul Rahman Zaidah[3,4]

Affiliations: [1]MPath (Microbiology) Student, Department of Medical Microbiology & Parasitology, School of Medical Sciences, Universiti Sains Malaysia, Kubang Kerian, 16150 Kelantan, Malaysia; [2]Infectious Disease Physician, Department of Medicine, School of Medical Sciences, Universiti Sains Malaysia, Kubang Kerian, 16150 Kelantan, Malaysia; [3]Hospital USM, Health Campus, USM, 16150 Kubang Kerian, Kelantan, Malaysia; [4]Medical Microbiologist, Department of Medical Microbiology & Parasitology, School of Medical Sciences, Universiti Sains Malaysia, Kubang Kerian, 16150 Kelantan, Malaysia.

Corresponding Author: Abdul Rahman Zaidah, Department of Medical Microbiology & Parasitology, School of Medical Sciences, Universiti Sains Malaysia, Kubang Kerian, 16150 Kelantan, Malaysia; Email: drzaidah@usm.my

Received: 27 June 2019
Accepted: 08 August 2019
Published: 13 September 2019

INTRODUCTION

Burkholderia pseudomallei is a highly pathogenic, Gram-negative saprophyte found in soil, groundwater, stagnant streams, rice paddies, and ponds [1]. Melioidosis is regularly reported from Southeast Asia and Northern Australia as well as from South Asia, China, and Brazil [2]. The most common risk factor identified was diabetes mellitus. Others include exposure to contaminated water or soil and occupation-related activities. Majority of patients presented with pneumonia which counted for 36–51% of total cases. Other less common sites of infection include blood stream, skin, bone, soft tissue, and liver [2–4]. Melioidosis presented with acute cholangitis is uncommon [5]. In this case report, we would like to share an atypical presentation of melioidosis.

CASE REPORT

A 45-year-old Malay lady with hypertension and asthma was a referred case from a district hospital for further management of acute obstructive cholangitis. She presented with generalized colicky abdominal pain for two weeks which later became persistent. It was associated with loss of appetite, nausea, and significant weight loss. Four days prior to admission she had high grade fever associated with chills and rigor. Otherwise denied symptoms such as tea colored urine or pale-colored stool. At the Emergency Department, the patient was alert, conscious, jaundiced, afebrile with a blood pressure of 104/70 mmHg and a heart rate (HR) of 70 bpm. Lung findings were unremarkable. There was generalized epigastric and right hypochondriac tenderness upon abdominal examination. She was then admitted to a general surgical ward with a diagnosis of acute cholecystitis for further management.

However, just a few hours after admission, she deteriorated when her blood pressure dropped to 99/58 mmHg and HR of 140 bpm. At the same time, she developed respiratory distress with respiratory rate (RR) of 35–40 bpm and oxygen saturation (SpO_2) of 88% on high flow mask. Arterial blood gases (ABGs) were taken urgently and showed type I respiratory failure with metabolic acidosis. She was electively intubated and was transferred to the intensive care unit (ICU) for further management.

Other investigations showed mild renal impairment. Urea was 13.2 mmol/L, sodium 126 mmol/L, potassium 3.0 mmol/L, and creatinine 325 umol/L. Total white cell count (TWC) was 15.62×109/L with hemoglobin (Hb) of 9.3 g/dL and normal platelets count (196×109/L) on full blood count (FBC) analysis. There was markedly increased of alkaline phosphatase (ALP), 899 µ/L and aspartate transaminase (AST), 64 µ/L. She was diagnosed with diabetes mellitus type 2 during this admission when her HbA1c was high (13.3%). Chest and abdominal radiographs showed clear lung fields and normal bowel outline (bowel not dilated). Based on the clinical presentation and investigations results, she was treated empirically as septicemia secondary to ascending cholangitis with intravenous (IV) meropenem 1 g eight hourly and IV metronidazole 500 mg eight hourly.

Emergency ultrasonography of hepatobiliary system was done on the day of admission and showed hepatosplenomegaly as well as splenic lesion suggestive of an early abscess. Computed tomography scan of the abdomen was done two days later since the patient was not stable for the procedure to be done much earlier. Findings showed features of multifocal splenic and liver abscesses. Initial blood culture taken on the day of admission grew coagulase-negative *Staphylococcus* which was regarded as a contaminant. Repeat blood culture five days later grew two types of Gram-negative bacteria, lactose fermenter, and nonlactose fermenter organisms with metallic sheen colonies (Figure 1). The bacteria were later confirmed as *B. pseudomallei* by automated VITEK2 identification system with 99% probability. She was then treated as disseminated melioidosis with high dose of IV ceftazidime, 2 g every eight hourly.

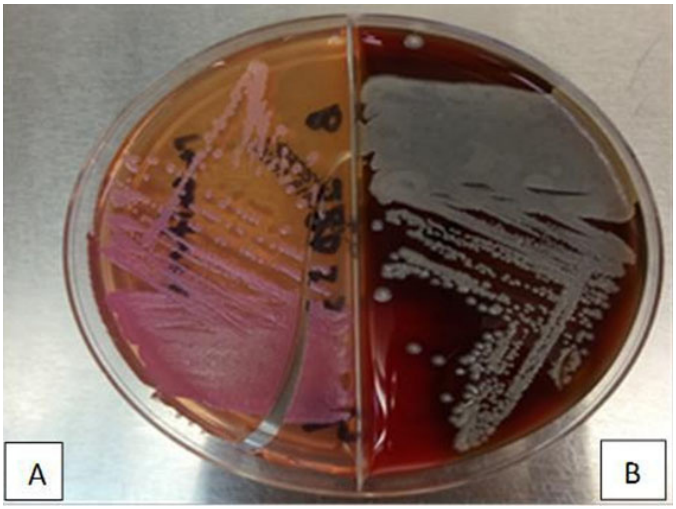


Figure 1: Cultured plate showing two types of Gram-negative bacteria isolated from the patient's blood. (A) Lactose fermenter (LF) organism on MacConkey agar (*Escherichia coli*). (B) Cream-colored colonies with a metallic sheen on blood agar, later identified as *B. pseudomallei*.

She was managed in the ICU for eight days, responded well to the melioidosis regime and was able to be transferred to a general ward to complete the IV antibiotics. Repeat ultrasonography of hepatobiliary system after two weeks of antibiotics therapy showed the presence of splenic abscess with resolved liver abscess. She then continued with IV ceftazidime for four weeks and discharged with oral trimethoprim-sulfamethoxazole to be completed for a total of five months.

DISCUSSION

Melioidosis can present with varies clinical manifestations, from acute to subacute or chronic diseases. These nonspecific presentations warrant high clinical suspicion, especially in endemic area since mortality rate is high if treatment is delayed. Studies from Australia and Thailand documented case fatality between 19% and 36% of their cases [6, 7]. Basically, *B. pseudomallei* is acquired through direct contact with the contaminated soil or water, inhalation, and ingestion. However, disease severity and duration of incubation period are associated with the size of infective inoculum [8]. Incubation period varies widely from case to case. A patient was reported to manifest the signs and symptoms of the disease after 62 years of exposure [9].

The clinical patterns of cases reported from Malaysia were consistent with cases from other countries in South and Southeast Asia. They shared common primary presentations of pneumonia and soft tissue abscesses, while diabetes as a major risk factor [2]. Among the cases,

pneumonia was the most common disease manifestation followed by soft tissue abscess. Mortality was three-fold higher in pneumonia compared to soft tissue abscess [2]. Internal abscesses were mainly noted in subcutaneous tissue followed by liver, spleen, and lung were the third most common sites [2]. During preparing of this manuscript, only two cases of melioidosis presented with cholangitis/cholecystitis had been reported [5, 10]. The first case is a Singaporean man presenting with fever, upper abdominal pain, and yellow eyes. The second case is a middle-aged man from Malaysia who presented with typical signs and symptoms of cholangitis; fever, jaundice, and right hypochondriac pain. This current third case was admitted with similar signs and symptoms of acute cholangitis. Acute abdomen is not a common presentation of melioidosis despite the fact that liver abscesses are among the common clinical manifestations [11].

Diagnosis for this patient was confirmed five days after admission since clinical presentations were pointing toward another diagnosis. Furthermore, she has no background history of diabetes mellitus, a major risk factor and initial peripheral blood culture grew skin contaminant. Repeat blood cultures confirmed the diagnosis. Isolation of *B. pseudomallei* from clinical specimens is still considered to be the "gold standard" for the diagnosis. The culture method has high specificity (100%) but low in sensitivity (60%) [12]. It depends on the type of samples, culture media used, and method of identification. *Burkholderia pseudomallei* is a motile aerobic, nonspore forming Gram-negative bacillus. In the laboratory, it grows aerobically on most agar media and produces clearly visible colonies within 24–48 hours at 37°C. Ashdown medium or modifications of it are commonly used and the organism demonstrates differing colonial morphology, with mostly smooth colonies initially and dry or wrinkled colonies on further incubation and can be identified reliably from its biochemical profile with kit-based systems [13]. Various serological test are available, however, these tests are of low sensitivities and specificities or lack of standardizations [14].

Timely identification of *B. pseudomallei* as offending organism is very important because it is intrinsically resistant to many commonly used antibiotics. Appropriate antibiotic therapy for melioidosis will improve the prognosis in this highly fatal, disseminated, and bacteremic form of infection. All patients with *B. pseudomallei* infection should be treated with initial intensive therapy followed by eradication therapy. In the intensive therapy, IV ceftazidime, meropenem, or imipenem need to be given for at least two to four weeks duration [15]. The following eradication therapy is given for a duration of 12–20 weeks with oral trimethoprim-sulfamethoxazole or amoxicillin/clavulanate potassium for those who cannot tolerate trimethoprim-sulfamethoxazole [15].

High suspicion for melioidosis is needed in a high-risk group of patient, such as the patient with diabetes mellitus, especially in an endemic region for melioidosis, in view of atypical presentations of the disease and also high mortality rate associated with it. The possibility of melioidosis as the infective etiology in patients presenting with severe ascending cholangitis or acute cholecystitis, particularly in susceptible patients and in endemic countries should be considered. Early recognition and prompt treatment with appropriate antibiotics would be lifesaving.

CONCLUSION

Acute cholangitis or cholecystitis is a rare presentation of melioidosis. High degree of suspicion is very crucial in a patient presented with any severe illness especially in an endemic area. Patients who are at risk for diabetes mellitus should be retested to confirm the current status especially when they presented with acute severe illness since early diagnosis and appropriate antibiotics management are the key factors for survival in melioidosis.

****

Acknowledgments

We thank the Management of the Hospital Universiti Sains Malaysia (USM), Kubang Kerian, Kelantan for granting the permission to the investigators to use patients' medical record; space and assets belong to the hospital during the process of conducting the research.

Author Contributions

Syazwani Mohd Tajudin – Conception of the work, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Alwi Muhd Besari – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Nabilah Ismail – Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Siti Asma' Hassan – Conception of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be

published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Abdul Rahman Zaidah – Conception of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission
The corresponding author is the guarantor of submission.

Source of Support
None.

Consent Statement
Not applicable. All possible efforts have been made to protect the identity of the patient.

Conflict of Interest
Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.

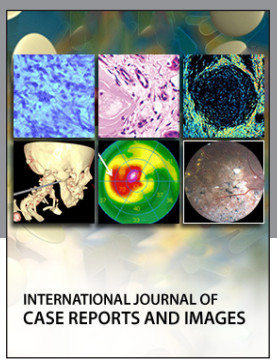
Copyright
© 2019 Syazwani Mohd Tajudin et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

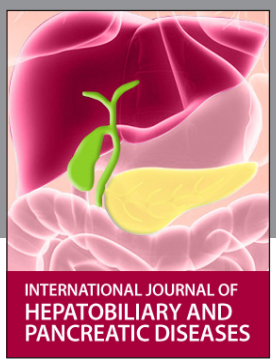
REFERENCES

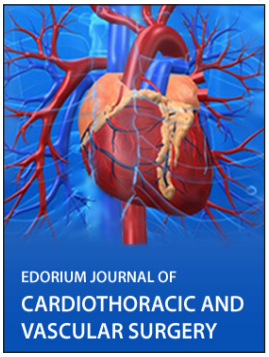
1. Rattanavong S, Wuthiekanun V, Langla S, et al. Randomized soil survey of the distribution of *Burkholderia pseudomallei* in rice fields in Laos. Appl Environ Microbiol 2011;77(2):532–6.
2. Kingsley PV, Leader M, Nagodawithana NS, Tipre M, Sathiakumar N. Melioidosis in Malaysia: A review of case reports. PLoS Negl Trop Dis 2016;10(12):e0005182.
3. Meumann EM, Cheng AC, Ward L, Currie BJ. Clinical features and epidemiology of melioidosis pneumonia: Results from a 21-year study and review of the literature. Clin Infect 2012;54(3):362–9.
4. Zueter A, Yean CY, Abumarzouq M, Rahman ZA, Deris ZZ, Harun A. The epidemiology and clinical spectrum of melioidosis in a teaching hospital in a North-Eastern state of Malaysia: A fifteen year review. BMC Infect Dis 2016;16:333.
5. Ye QJ, Desai SR, Tan EK. A case of melioidosis presenting as acalculous cholecystitis. Cureus 2018;10(6):e2864.
6. Currie BJ, Fisher DA, Howard DM, et al. Endemic melioidosis in tropical northern Australia: A 10-year prospective study and review of the literature. Clin Infect Dis 2000;31(4):981–6.
7. Bhengsri S, Baggett HC, Jorakate P, et al. Incidence of bacteremic melioidosis in eastern and northeastern Thailand. Am J Trop Med Hyg 2011;85(1):117–20.
8. Cheng AC, Currie BJ. Melioidosis: Epidemiology, pathophysiology, and management. Clin Microbiol Rev 2005;18(2):383–416.
9. Ngauy V, Lemeshev Y, Sadkowski L, Crawford G. Cutaneous melioidosis in a man who was taken as a prisoner of war by the Japanese during World War II. J Clin Microbiol 2005;43(2):970–2.
10. Mohamad N, Ponnusamy S, Devi S, Manikam R, Idrus II, Bakar NHA. Melioidosis in acute cholangitis of diabetic patient: A forgotten diagnosis. Res Rep Trop Med 2012;3:103–6.
11. Nathan S, Chieng S, Kingsley PV, et al. Melioidosis in Malaysia: Incidence, clinical challenges, and advances in understanding pathogenesis. Trop Med Infect Dis 2018;3(1). pii: E25.
12. Limmathurotsakul D, Jamsen K, Arayawichanont A, et al. Defining the true sensitivity of culture for the diagnosis of melioidosis using Bayesian latent class models. PLoS One 2010;5(8):e12485.
13. Puthucheary SD. Melioidosis in Malaysia. Med J Malaysia 2009;64(4):266–74.
14. Lau SK, Sridhar S, Ho CC, et al. Laboratory diagnosis of melioidosis: Past, present and future. Exp Biol Med (Baywood) 2015;240(6):742–51.
15. Wiersinga WJ, Currie BJ, Peacock SJ. Melioidosis. N Engl J Med 2012;367(11):1035–44.

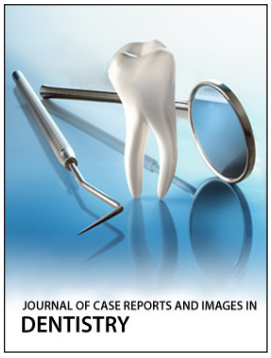
Access full text article on other devices

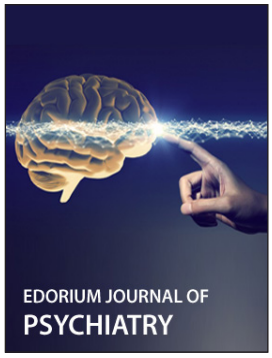
Access PDF of article on other devices

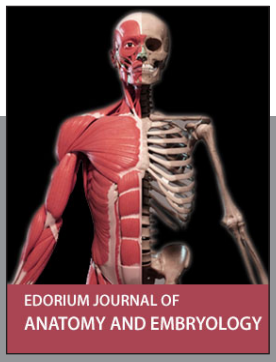


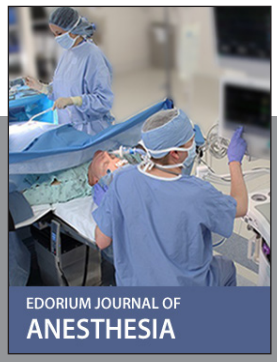












Submit your manuscripts at

www.edoriumjournals.com