

Unusual Presentations of Abdominal Melioidosis

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Abstract

Melioidosis is an endemic bacterial infection caused by soil saprophyte, *Burkholderia pseudomallei*. It infects adults with risk factors or immunosuppressed and exposed to moist soil. Its significance lies in its varied clinical presentation and high mortality (40%). We present two cases of abdominal melioidosis with unusual clinical presentations. The first case presented with intractable hiccups and had isolated splenic melioidosis with contained rupture. The second case presented with fever and acute abdominal pain found to have pancreatic melioidosis and splenic vein thrombosis. Both the patients were treated with IV antibiotics and subsequently discharged after improvement in symptoms. The imaging findings of isolated type of melioidosis can mimic various other infections and granulomatous disease. Hence high index of clinical suspicion for patients presenting from endemic areas will narrow down the differential diagnosis.

Keywords: *Burkholderia* infection, melioidosis, splenic abscess

INTRODUCTION

Melioidosis, also called “Whitmore’s disease,” was first reported in Myanmar in 1912.^[1] It is highly prevalent in Southeast Asia, and is caused by a motile, aerobic Gram-negative bacillus called *Burkholderia pseudomallei*.^[2] It affects adults with diabetes mellitus (37%–60%), renal disease, and alcoholism and those immunosuppressed as a result of either disease or drugs.^[2] However, it has no association with HIV infection.^[3] The organism is transmitted through abraded skin defect when exposed to moist soil and rarely by inhaling or ingesting the causative agent. The presentation can be varying from subclinical to fulminating disease with multiple organ involvement and even leads to mortality. It is categorized as a Class II bioterrorism agent.^[4] Lung is the most commonly involved organ. The most common extrapulmonary organs to be involved are the spleen and liver, followed rarely by the pancreas, prostate, bones, and skeletal muscle.^[3] Although radiological findings are characteristic, delayed diagnosis is common in isolated single organ involvement and the radiological features can mimic various other infections and granulomatous disease. Hence, the diagnosis of melioidosis requires clinical suspicion in addition to the imaging findings. Confirmation of the diagnosis is usually microbiological. Here, we report two cases of melioidosis with unusual clinical presentations.

CASE REPORT

Case 1

A 47-year-old green-grocer, native of Assam, presented with 10-month history of intractable hiccups, associated with low-grade fever, bloating, and weight loss. He was a chronic diabetic, an active smoker, and an alcoholic. His abdominal examination was normal. His glycemic control was suboptimal (postprandial blood sugar – 300 mg/dL and hemoglobin A1c [HbA1c] 9.4%), and other blood investigations were within normal limits. Chest radiograph was normal. Ultrasonography of the abdomen showed multiple hypoechoic lesions with mild splenomegaly. He was further evaluated with contrast-enhanced computed tomography (CECT) abdomen which revealed splenomegaly with multiple, small, ring enhancing lesions (6–8 mm). A few of these lesions showed a clustered pattern of distribution. A thin rim of perisplenic fluid was noted near the subcapsular lesions, suggesting contained rupture [Figure 1a]. Rest of the abdominal organs was

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unremarkable. Magnetic resonance imaging (MRI) abdomen showed multiple T2W hyperintense splenic lesions with T2W hypointense rim [Figure 1b]. Diffusion-weighted imaging showed restricted diffusion in these lesions, suggesting abscesses [Figure 1c]. He underwent an ultrasound-guided, fine-needle aspiration which yielded a few milliliters of purulent fluid. The pus culture revealed safety pin appearance of Gram-negative bacilli in the Gram staining and metallic sheen on MacConkey’s agar plate. *B. pseudomallei* was identified from the aspirate of the splenic abscess, using the VITEK2 with updated Advanced Expert System (AES) [Biomerieux, Marcy l’Etoile, France] automated system after an incubation period of 72 h. The patient was therapeutically managed with intravenous ceftazidime (2 g every 6 h) for 2 weeks and subcutaneous insulin till he achieved his glycemic control. The patient showed gradual improvement in hiccups symptoms and was subsequently discharged and suggested for follow-up.

Case 2

A 62-year-old male from Tamil Nadu presented with a 2-week history of fever and acute onset of abdominal pain for 4 days. He was a chronic diabetic and hypertensive and not on proper medications. Physical examination showed a pulse rate of 105 beats/min and blood pressure of 148/90 mmHg. Abdominal examination revealed tenderness in the left hypochondrium and epigastrium. Examination of other systems was unremarkable. His blood investigations showed normal hemogram, serum amylase, and lipase levels. His liver and renal function tests were within normal limits. His HbA1c was 9.2%, suggesting poor glycemic control. CECT abdomen revealed mildly bulky distal body and tail regions of pancreas with peripancreatic fat streakiness [Figure 2a]. The retro-pancreatic portion of the splenic vein was distended with non-enhancing intraluminal thrombus and vessel wall enhancement, suggestive of acute thrombosis [Figure 2b]. Prominent venous collaterals were noted in the periportal, peripancreatic, omental, and retroperitoneal locations. The head, neck, and proximal body of the pancreas were normal. The main pancreatic and common bile ducts were not dilated. Mild splenomegaly was also noted. MRI abdomen showed a mildly bulky pancreas. The retro-pancreatic portion of the splenic vein showed T2 hyperintense thrombus with restricted diffusion [Figure 2c]. In view of the history of fever with normal amylase and

lipase levels, the possibility of infective cause of pancreatic inflammation with splenic vein thrombosis was postulated.

Blood culture from this patient revealed a safety pin appearance of Gram-negative bacilli in the routine Gram staining [Figure 3a], and colonies showed a metallic sheen on MacConkey’s agar plate [Figure 3b]. After an incubation period of 72 h, using the VITEK2 (Biomerieux) automated system, the causative agent was revealed to be *B. pseudomallei*. The patient was managed with parenteral meropenem (1 g every 8 h t.i.d) and oral co-trimoxazole (960 mg every 12 h b.i.d) with subcutaneous enoxaparin (1 mg/kg b.i.d) for splenic vein thrombosis. He received subcutaneous antidiabetic medication human Actrapid® for 2 weeks. He improved symptomatically during the intensive phase of treatment and achieved glycemic control, subsequently discharged after switching to oral anticoagulation and oral hypoglycemic drugs, and suggested for regular follow-up.

DISCUSSION

Melioidosis is an emerging infective disease caused by *B. pseudomallei*. Globally, melioidosis is endemic in northern Australia, Thailand, Singapore, and Malaysia. In India, higher incidence of cases is seen in western and eastern coastal belts, with maximum cases reported from cities such as Mangalore, Chennai, Puducherry, West Bengal, and Assam.^[3,5] Males are at higher risk due to occupational exposure (male: female = 1.4:1).^[3] Abdominal melioidosis has a diverse clinical presentation depending on the organ involved by the organism. Spleen is the most frequently involved abdominal organ followed by liver.^[3,6] It commonly presents as hepato-splenic abscesses and the concurrent involvement of liver and spleen is more likely to be associated with melioidosis than with any other infections.^[3] The affected spleen is often clinically enlarged and mildly tender. Acute or chronic presentation of the spleen occurs by hematogenous route. The acute form may involve multiple organs, and splenic involvement is usually incidental. The chronic form can have isolated splenic involvement, and patients may not exhibit any symptoms.^[7] On imaging, multiple, discrete lesions (varying from 0.5 to 1.5 cm) are usually seen in the spleen. Subcapsular collections with or without perisplenic extensions are common.^[8]

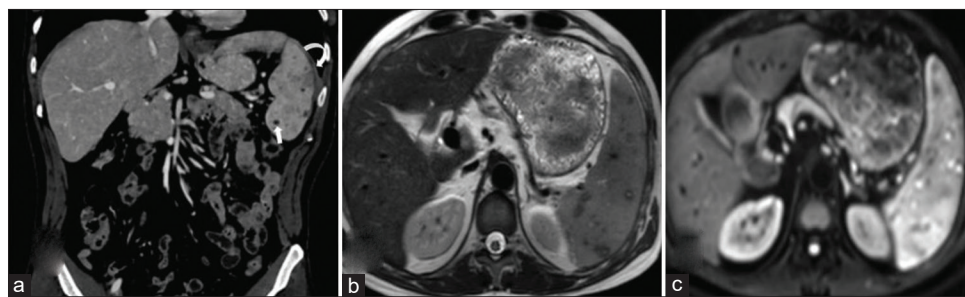


Figure 1: (a-c) Contrast-enhanced computed tomography abdomen, coronal section showing (a) multiple, tiny hypodense splenic lesions (thin arrow) with clustered pattern of distribution in subcapsular location and perisplenic extension (curved arrow). Magnetic resonance imaging abdomen, axial section showing T2-weighted image (b) and diffusion-weighted imaging (c) showing multiple T2 hyperintense lesions with diffusion restriction in the spleen

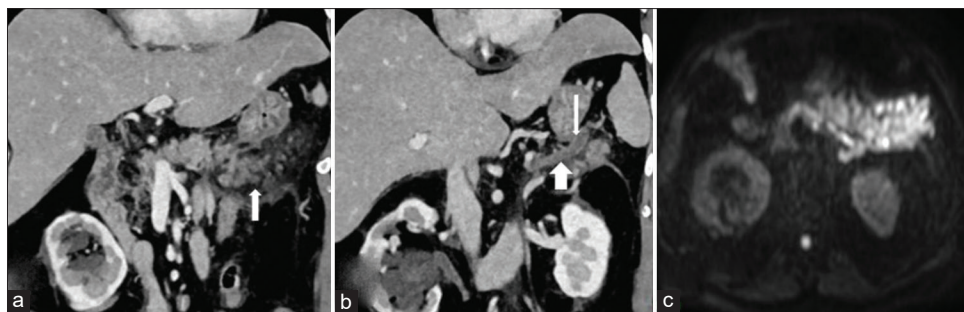


Figure 2: (a-c) Contrast-enhanced computed tomography abdomen, coronal section showing (a) heterogeneous hypoenhancement in the tail of the pancreas (arrow). (b) Nonenhancing thrombus involving the splenic vein (thick arrow) from the confluence till the intraparenchymal branches with enhancing thickened wall of the splenic vein (thin arrow). (c) Magnetic resonance imaging abdomen, axial section showing diffusion restriction with corresponding low apparent diffusion coefficient in the distal body of the pancreas and in the splenic vein thrombus

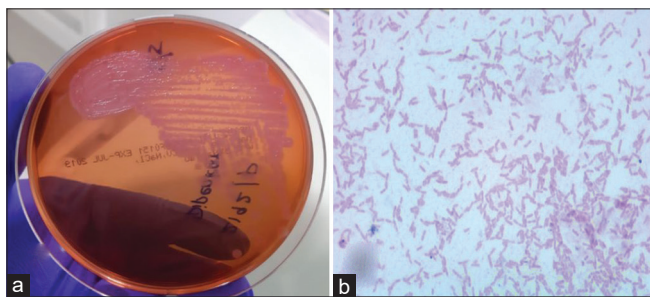


Figure 3: (a and b) Bacterial colony morphology of pus and blood culture. (a) MacConkey agar showing typical nonlactose-fermenting, metallic sheen appearing colonies. (b) Gram smear revealing characteristic bipolar Gram-negative bacilli ($\times 100$)

Our first case was from an endemic area of melioidosis (one of the states from the northeast of India – Assam) and has multiple comorbid conditions, namely a chronic diabetic, an active smoker, and a chronic alcoholic. Because he is a green-grocer by occupation, contact with contaminated soil could be the most possible source of exposure. The presentation was chronic and was due to rupture of splenic abscess in the absence of local peritoneal inflammation. Previously reported cases of splenic abscess rupture^[9-11] had acute presentation with associated peritonitis. In our case, there were multiple, small peripheral abscesses in the diaphragmatic surface of the spleen. These abscesses ruptured but were well contained and did not directly communicate with the peritoneal cavity, leading to the absence of local peritoneal inflammation. This could be the reason for a chronic presentation in this case. The above patient also presented with the novel symptom of “intractable hiccups,” a feature as of yet never been described in literature as a chief complaint. The subdiaphragmatic location of an abscess leads to irritation of the diaphragm which is supplied by phrenic nerve, causing this chronic symptom of hiccups. The possible differential diagnosis for the clinical and imaging features is tuberculosis, brucellosis, and fungal. There is no specific clinical feature to suggest melioidosis. As his clinical presentation was low-grade fever and chronic weight loss with intractable hiccups, the imaging features of multiple, small splenic abscesses, some of which showed a contained rupture along the superolateral surface of the spleen irritating

the diaphragm, could explain the above symptoms. Thus, in an endemic area, splenic melioidosis with contained rupture should be considered in the differential for patients with known risk factors and presenting with intractable hiccups.

Our second case was also from an endemic area (state of Tamil Nadu), but he had poor glycemic control, predisposing to melioidosis. The most possible source for acquiring infection is through ingestion of contaminated water. Pancreatic involvement by melioidosis has been rarely reported. The imaging findings of pancreatic melioidosis include a bulky pancreas with focal large abscess or multifocal pancreatic micro-abscesses.^[3,12] Other associated findings are splenic vein thrombosis and peripancreatic fat-stranding. Typically, these patients have normal amylase and lipase levels.^[12] The imaging revealed mild interstitial pancreatitis with no evidence of abscess formation. This case had the distinction of manifesting with septic splenic vein thrombosis with mild interstitial pancreatitis in the absence of pancreatic abscess. To the best of our knowledge, all the reported cases of pancreatic melioidosis in the literature had abscesses in pancreatic parenchyma.^[12] We suggest the inclusion of a diagnosis of “pancreatic melioidosis” in high-risk patients from an endemic area, who present with fever and pain abdomen with imaging features of interstitial pancreatitis with septic splenic vein thrombosis in spite of the normal levels of serum amylase and serum lipase levels, as seen in our case. The absence of pancreatic abscess should not necessarily exclude the above diagnosis. The diagnosis was confirmed with blood culture in our case. Clinicians and radiologists should be aware of this rare presentation of pancreatic melioidosis, where it can present as interstitial pancreatitis and without abscess formation.

The culture and isolation of *B. pseudomallei* from the clinical aspirate is considered the confirmatory diagnosis for melioidosis.^[13] At our institution, we always initiate high dose of intensive-phase antibiotics: combination of intravenous ceftazidime (1–2 g tid.) and amoxicillin/clavulanic acid (1.2–2.4 g tid.) as a drug of choice or monotherapy (meropenem 1–2 g tid.) for 4–8 weeks and eradication phase of antibiotics: oral co-trimoxazole (960 mg bid) or oral doxycycline 200 mg daily, given 6 months for melioidosis patients with underlying risk factors presenting as sepsis.^[14] In our experience, all

visceral involvement of melioidosis regressed with the above antibiotics. Hence, improving awareness among the high-risk group patients with diabetes and outdoor labor will reduce the mortality and morbidity rate of melioidosis.

CONCLUSION

Isolated multiple splenic abscesses with subcapsular collection in an adult with underlying comorbidities and a history of visit to an endemic area should raise the suspicion of melioidosis. Similarly, pancreatic and peripancreatic inflammation with splenic vein thrombosis and normal amylase and lipase levels should raise a suspicion of pancreatic melioidosis. It is important for the clinicians working in nonendemic areas to be aware of this diagnosis and include it in their list of differential diagnoses.

Research quality and ethics statement

The authors of this manuscript declare that this scientific work complies with reporting quality, formatting, and reproducibility guidelines set forth by the EQUATOR Network. We also certify that we have not plagiarized the contents in this submission and have done a plagiarism check.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for 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.

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