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Ruptured splenic abscess and splenic vein thrombosis secondary to melioidosis: A case report

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ABSTRACT

Rationale: *Burkholderia pseudomallei* is a Gram-negative bacterium and the causative pathogen of melioidosis, which manifests with a broad spectrum of clinical syndromes. Melioidosis is associated with high mortality and is endemic across tropical areas, especially in Southeast Asia and northern Australia.

Patient concern: A 24-year-old diabetic male complained of fever and left upper quadrant abdominal pain for one-week duration.

Diagnosis: Melioidosis with ruptured splenic abscess and splenic vein thrombosis.

Interventions: Antimicrobial therapy (intensive therapy: intravenous ceftazidime, eradication therapy: oral trimethoprim-sulfamethoxazole), and anti-coagulation (enoxaparin, then warfarin).

Outcomes: Resolution of splenic abscess and splenic vein thrombosis.

Lessons: Both splenic abscess and splenic vein thrombosis are uncommon but severe complications associated with melioidosis. Ultrasound is useful for diagnosis and monitoring response to treatment in such cases.

KEYWORDS: Melioidosis; Splenic abscess; Splenic vein thrombosis; *Burkholderia pseudomallei*

1. Introduction

Melioidosis, also known as “Whitmore disease”, is an infectious disease caused by *Burkholderia pseudomallei* (*B. pseudomallei*), and is endemic in northern Australia and Southeast Asia[1]. The clinical spectrum of melioidosis is complex, wide-ranging, and includes latent infection, local cutaneous lesions, subacute pneumonia, focal organ abscess, musculoskeletal infection, and lethal fulminant pneumonia[1,2]. The most common cause of splenic vein thrombosis is pancreatic diseases[3], including chronic pancreatitis, pancreatic

pseudocyst, pancreatic tumour, pancreatic abscess and acute pancreatitis[4]. Splenic vein thrombosis secondary to melioidosis is very rare and has only been described in a few case reports[5,6]. We reported a case of melioidosis with rare manifestations of splenic vein thrombosis and splenic abscess.

2. Case report

This study was supported by the National Medical Research Register of Malaysia (NMRR-20-193-53257). Written informed consent for publication of the clinical details was obtained from the patient. A 24-year-old male presented in June 2019 at Sarawak General Hospital, Sarawak, Malaysia with a one-week history of fever, poor appetite and left upper quadrant abdominal pain. He denied any symptoms of respiratory tract infection or reactivation of pulmonary tuberculosis. There was no history of gastrointestinal bleeding. His medical history included poorly controlled type I diabetes mellitus on a twice-daily premixed insulin regime with recent glycated haemoglobin (HbA1c) of 10% and a history of smear-positive pulmonary tuberculosis in 2015 for which he had undergone a 6-month anti-tuberculous treatment.

Upon arrival at the hospital, he was febrile and lethargic-looking. Physical examination revealed a blood pressure of 137/80 mmHg, pulse rate of 107 beats/min, and temperature of 38.2 °C. His

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respiratory rate was 18 breath/min and the oxygen saturation on room air measured by pulse oximetry was 98%. The abdomen was soft but tender at the left hypochondriac region. The remainder of the systemic examination was unremarkable. The chest radiograph did not reveal any obvious signs of pneumonia or pneumoperitoneum.

Haematological analysis showed haemoglobin of 12.1 g/dL (normal: 12.0-15.0 g/dL), white blood cell count of $13.5 \times 10^3/\mu\text{L}$ (normal: $4.0\text{-}10.0 \times 10^3/\mu\text{L}$) and platelet count of $220 \times 10^3/\mu\text{L}$ (normal: $150\text{-}410 \times 10^3/\mu\text{L}$). The renal, liver function tests and serum amylase were within the normal range. The arterial blood gas revealed good oxygenation and the absence of metabolic disturbance. Urgent abdominal ultrasonography revealed splenomegaly with the presence of multiple splenic abscesses (largest measuring 1.5 cm \times 5.9 cm \times 2.6 cm) and normal splenic vein thrombosis (Figure 1). Meanwhile, the pancreas was normal.

The blood culture yielded *B. pseudomallei*, and the time to detection was 1 d/3 h/30 min. Preliminary identification using Gram staining and then matrix-assisted laser desorption/ionization-time of flight mass spectrometry confirmed *B. pseudomallei*. The isolate was sensitive to amoxicillin-clavulanic acid, ceftazidime, and trimethoprim-sulfamethoxazole (Minimum inhibitory concentration=1.5 $\mu\text{g}/\text{mL}$). A confirmatory test by real-time PCR was not available in our setting.

He was administered with intravenous ceftazidime 2 g every 6 h as intensive phase treatment for melioidosis. Subcutaneous enoxaparin (1 mg/kg twice daily) was also started for the treatment of splenic vein thrombosis. In the meantime, he was referred to the endocrine team for blood sugar control optimization in view of multiple episodes of hyperglycemia in the ward.

In view of the large splenic abscess, the patient was referred for percutaneous drainage of the abscess. However, it was not feasible in view of technical difficulty and the abscess has not liquefied. Hence, it was decided for the continuation of antibiotic therapy and re-evaluation later by ultrasonography. He had a single isolated fever spike (38°C) on day 6 of hospitalization and subsequently afebrile. There was no sign of peritonism. Abdominal pain was gradually improved with analgesia.

After 2 weeks of treatment with intravenous ceftazidime, a repeat abdominal ultrasonography showed an unchanged size of splenic abscess which has ruptured, and persistent thrombosis of the splenic vein (Figure 2). Oral trimethoprim-sulfamethoxazole was added during the intensive-phase treatment. The patient remained afebrile and responded well to antibiotic therapy. Anti-coagulation was then switched to oral warfarin, adjusted based on the target international normalized ratio range of 2.0-3.0.

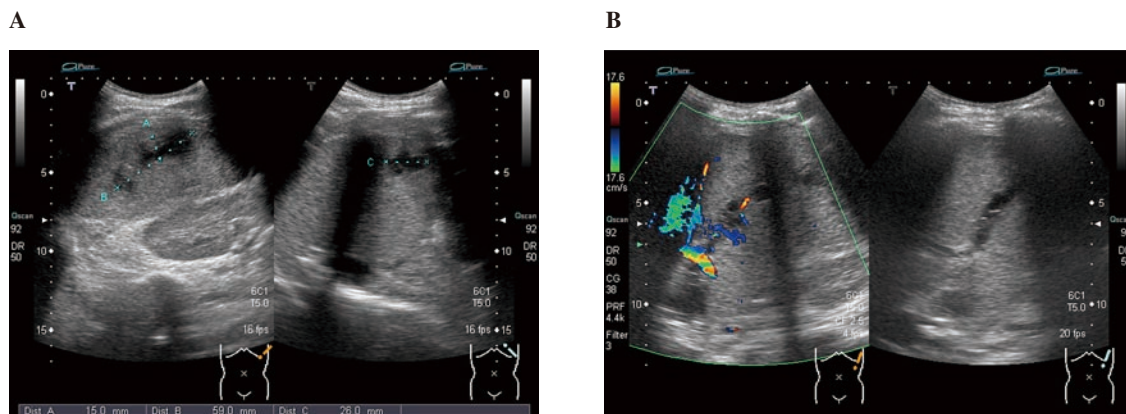


Figure 1. Urgent abdominal ultrasonography revealed splenomegaly with the presence of multiple splenic abscesses. A: Mixed echogenic collection in the spleen measuring 1.5 cm \times 5.9 cm \times 2.6 cm; B: Thrombosed splenic vein.

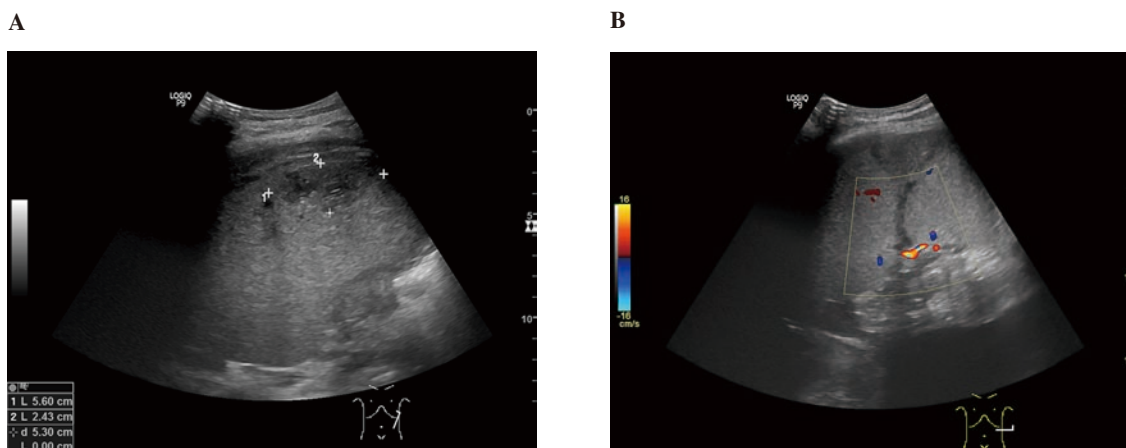


Figure 2. Repeat abdominal ultrasonography showed an unchanged size of splenic abscess which has ruptured, and persistent thrombosis of the splenic vein. A: Unchanged size of splenic abscess (5.6 cm \times 2.4 cm); B: Thrombosis of the splenic vessel branching into the splenic parenchyma and splenic hilum.

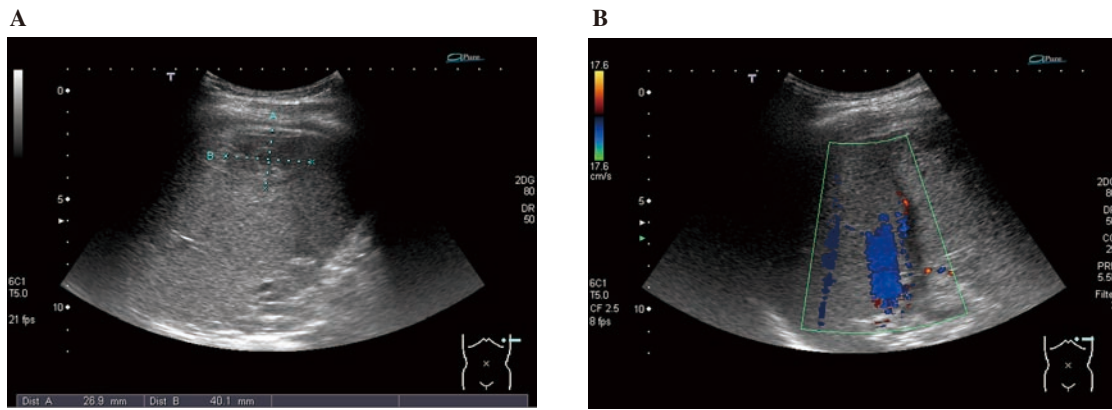


Figure 3. Abdominal ultrasonography showed smaller splenic abscess with residual splenic vein partial thrombosis. A: Reduced size of the splenic abscess (2.7 cm×4.0 cm); B: Residual splenic vein thrombosis.

At the one-month interval, abdominal ultrasonography was again repeated and showed smaller splenic abscess (largest, from previously 2.4 cm×5.6 cm to 2.7 cm×4.0 cm) with residual splenic vein partial thrombosis (Figure 3). Intravenous ceftazidime was given for a total of 6 weeks and oral trimethoprim-sulfamethoxazole was continued for 6 months. Ultrasound scan done at a third-month interval showed further reduction of the size of the splenic abscess and completely re-canalized splenic vein (Figure 4A). Hence, oral anti-coagulant was stopped after 3 months. Following treatment, the splenic abscesses eventually resolved at the end of treatment at 6 months (Figure 4B). During subsequent follow-up review, he remained well and showed no signs of disease recurrence.

4. Discussion

B. pseudomallei is the pathogen of melioidosis, an infectious disease that can virtually affect every organ in the body and present with diverse clinical manifestations. Melioidosis is associated with a high mortality rate because of the early spread of infection to the blood. In Malaysia, the case fatality rate of melioidosis is high despite advances in treatment, ranged from one-third to about half of the patients (33-54%)[7]. Diabetes mellitus, preexisting renal diseases, thalassaemia, and occupational exposure to soil and contaminated water were shown to be significant risk factors for melioidosis[1-2,7]. This patient has underlying diabetes mellitus which predisposed him to melioidosis infection. We postulated that he most probably acquired the infection by inhalation of dust or contaminated soil during his work at a road construction site.

There have been a few reported cases of melioidosis-associated splenic vein thrombosis. Saïdani *et al.* reported a rare manifestation of splenic vein thrombosis in a patient with bacteraemic melioidosis, pulmonary, and hepatic abscesses[5]. Splenic vein thrombosis was reported as an associated pancreatic finding in melioidosis cases besides pancreatic abscesses, peripancreatic inflammation, and peripancreatic fat streaking[6]. Thrombotic events in melioidosis infection could be the result of an inflammatory response to systemic *B. pseudomallei* infection, leading to depletion of the natural endothelial modulators protein C, protein S, and antithrombin[8]. Mesenteric angiography with venous phase imaging is the gold standard of diagnosis of splenic vein thrombosis. Ultrasound and computed tomography may identify splenic vein thrombosis but are most helpful in delineating concomitant upper abdominal pathology[3].

Splenic abscesses are not commonly associated with melioidosis, and its incidence varies across different geographical areas. Splenic abscesses were less common, which presented in 28 out of 540 patients (5%) with melioidosis in Darwin, Australia[2]. On the other hand, almost three-quarters of patients with melioidosis were found to have splenic abscesses in a Thai study[9]. In this present case, a drainage of splenic abscess was not feasible because of technical difficulty and the abscess has not liquefied.

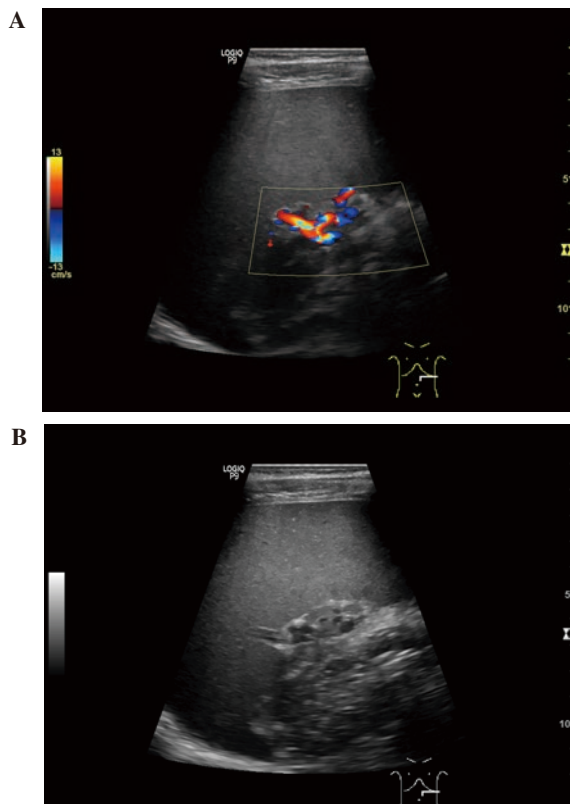


Figure 4. Abdominal ultrasonography at 3 month and 6 month. A: Recanalization of the splenic vein at 3 month. B: Resolution of the splenic abscess at 6 months.

Our patient received optimal antibiotic therapy consists of ceftazidime and trimethoprim-sulfamethoxazole, and subsequently improved clinically with the resolution of splenic abscesses and splenic vein thrombosis. In addition to antibiotic therapy, there are no conclusive recommendations about percutaneous splenic aspiration or splenectomy due to a lack of well-designed studies demonstrating the superiority of any therapy over others[10]. This implies that non-operative measures may be adequate in undrainable splenic abscesses secondary to melioidosis.

Melioidosis should be considered as a differential diagnosis in patients who present with splenic abscesses in an endemic area. Splenic vein thrombosis can be a potential complication. If appropriate treatment is not promptly instituted, there is a high risk of mortality.

Conflict of interest statement

The author reports no conflict of interest.

Authors' contribution

C.C.Y. managed the case and wrote the paper.

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