Case Report

**Burkholderia cepacia; an unusual cause of multiple splenic abscesses**

A case report

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Abstract

*Burkholderia cepacia* is an uncommon, multidrug resistant pathogen. We present a patient with a history of recent malignancy and uncontrolled diabetes, who presented with bacteraemia and multiple splenic abscesses due to this organism. She was managed conservatively, and made a good recovery. A high index of suspicion is required to arrive at the microbiological diagnosis and provide effective management.

**Keywords:** Burkholderia cepacia, multiple splenic abscesses, bacteraemia

Introduction

*Burkholderia cepacia* is an emerging bacterial pathogen, being mostly isolated from patients who have compromised immunity, or of nosocomial origin. Patients with cystic fibrosis (CF) and chronic granulomatous disease (CGD) are those most often affected by this uncommon pathogen. Even then, bacteraemia is an uncommon presentation. Infection with this organism is complicated by difficulties in microbiological diagnosis and multiple antibiotic resistance. We present a case of *Burkholderia cepacia* bacteraemia in a patient without CF or CGD, who developed multiple splenic abscesses.

Case Report

A 56-year-old female presented to the medical casualty, complaining of nausea, vomiting, difficulty in breathing and progressively deteriorating level of consciousness. She was afebrile. Her glycaemic control was poor. She had undergone right mastectomy in the recent past for carcinoma of the breast.

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After initial assessment, she was managed in the intensive care unit (ICU) as for septic shock complicated with diabetic ketoacidosis. Fluid resuscitation and inotropic support was given and intravenous (IV) meropenem 1 g 8 hourly and levofloxacin 500 mg twice daily was started empirically after obtaining blood and urine cultures.

Initial assessment showed neutrophil leukocytosis and elevated C-reactive protein. Abdominal ultrasound scan showed right sided acute pyelonephritis.

Despite antibiotics there was no improvement, with continuing high spiking fever even after 14 days. Three blood cultures taken sequentially and a urine culture were negative. Changing antibiotics to cefuroxime 750 mg 8 hourly, piperacillin-tazobactam 4.5 g 8 hourly and amikacin 500 mg twice daily and even antifungal fluconazole 400 mg daily had no effect.

After 13 days in the ICU, the fourth blood culture yielded a non-lactose fermenting, oxidase positive organism, identified as probable Pseudomonas species, on final subculture on day seven. The isolate was sensitive to ceftazidime and meropenem but resistant to gentamicin, amikacin and netilmicin. In total, three blood cultures drawn on ICU days 13, 15 and 17 were positive for the same organism with identical sensitivity pattern. Two of the three cultures (the first and the third) became positive on final subculture while one was positive after 24 hours of incubation. Since there was no clinical response to meropenem, antibiotics were changed to IV ceftazidime at 2 g 8 hourly and metronidazole 500 mg 8 hourly.

As these isolates lacked the typical culture characteristics of P. aeruginosa and invasive melioidosis was a differential diagnosis, the isolate and a sample of serum was sent to the Faculty of Medicine, Colombo for confirmation of melioidosis, of which the antibody level by indirect haemagglutination test, was elevated at 1: 640. However, real time-PCR for Burkholderia pseudomallei performed on the isolate was negative. Therefore, it was decided to send the isolate for automated biochemical system identification by Vitek 2 (bioMérieux).

Meanwhile, the patient’s fever continued to spike. As intra-abdominal pus collection was still suspected, contrast enhanced CT scan of abdomen was performed which showed multiple, small splenic abscesses.

The blood culture isolate was finally identified as Burkholderia cepacia, with a very good identification rating by Vitek 2 system. The patient’s fever responded after continuing ceftazidime for two weeks. The follow-up blood cultures were negative. She was sent to the ward after a month-long ICU stay, which was complicated by demyelinating polyneuropathy which improved with IV immunoglobulin and physiotherapy. Ceftazidime was continued for further four weeks and omitted. The repeat CT scan showed resolution of the abscesses.
Discussion

This case highlights several important issues.

The patient’s initial presentation was consistent with sepsis due to probable acute pyelonephritis. This may have responded to the initial antibiotics she received. *Burkholderia cepacia* infection which caused the prolonged fever may have been acquired nosocomially.

*Burkholderia cepacia* infection is uncommon and is usually seen in association with cystic fibrosis and chronic granulomatous disease. Outside of this population, risk factors are less well established. They include indwelling vascular catheters, abdominal surgery, malignancy and nosocomial transmission via the hands of healthcare workers and contaminated equipment. Our patient had uncontrolled diabetes and recent malignancy. She was intubated and mechanically ventilated soon after admission. These factors may have contributed to the dissemination and ultimate seeding of the organism in the spleen.

Splenic abscesses are uncommon and may arise due to bacteraemia, contiguous disease with local extension, splenic trauma, haemoglobinopathies and in intravenous drug abusers. Multiple small splenic abscesses are usually caused by bacteraemia. Most Asian studies have found *Klebsiella pneumoniae* as the predominant pathogen, while in studies done in the west, *Staphylococcus aureus* was the commonest isolate. To our knowledge, this is the first published case of multiple splenic abscesses due to *B. cepacia* in a non-cystic fibrosis patient.

Meliodosis caused by the related organism *B. pseudomallei* can also cause multiple splenic abscesses, in areas where the disease is endemic. Since a recent surveillance program found
nation-wide distribution of invasive melioidosis in Sri Lanka\textsuperscript{6} it was considered in the differential diagnosis of this patient. Melioidosis antibodies are considered positive depending on the endemicity of the region concerned. Per published literature, there may be cross reactivity between antibodies to lipopolysaccharide in the cell wall between \textit{B. pseudomallei} and other Gram-negative bacteria, such as \textit{B. cepacia}.\textsuperscript{7} However, the titres obtained in this instance are significant. Another possible scenario is a co-infection with \textit{B. cepacia} and \textit{B. pseudomallei}, which may account for the elevated melioidosis antibody titres.

The offending organism is usually isolated by culture of the drained abscess fluid. Blood cultures are positive in only about 50\% of cases. CT scanning is a useful tool for diagnosis.\textsuperscript{8} Our patient had persistent low grade bacteraemia that probably led to haematological seeding in the spleen, but the primary source could not be identified. It is also possible that the splenic abscesses were already present at the time of initial presentation, but were undetected due to the poor sensitivity of the ultrasound scan.

In conclusion, we report a case of \textit{B. cepacia} bacteraemia with multiple splenic abscesses, in a patient with uncontrolled diabetes and history of malignancy. She was managed conservatively and made a good recovery.

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**Conflict of interests:** None

**References**