Multidrug-resistant *Burkholderia cepacia* bacteremia in an immunocompetent adult diagnosed with dengue and scrub coinfection: A rare case report

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**ABSTRACT**

*Burkholderia cepacia* is an opportunistic nosocomial pathogen causing infections in immunocompromised hosts. Infection by *Burkholderia* in an immunocompetent host is a rare entity. We report a case of dengue and scrub coinfection complicated by *B. cepacia* bloodstream infection along with literature review of such infections in immunocompetent adults. Before the introduction of automated technologies, it was difficult to differentiate this organism from other aerobic Gram-negative nonfermenters, which have different intrinsic resistance profiles. Furthermore, *Burkholderia* has intrinsic as well as acquired resistance to various antimicrobials but not commonly to ceftazidime. To our knowledge, ours is the first case of multidrug-resistant *B. cepacia* infection in an immunocompetent host to be reported till date.

**Key Words:** Bacterial infection, *Burkholderia cepacia*, dengue fever

**INTRODUCTION**

*Burkholderia cepacia* is a rare opportunistic nosocomial pathogen causing infections in immunocompromised hosts such as those having cystic fibrosis, chronic granulomatous disease, hematological malignancies, chronic renal failure, and uncontrolled diabetes mellitus. In these patients, cepacia syndrome may present with fever, leukocytosis, pulmonary infiltrates, respiratory distress (necrotizing pneumonia), and septicemia. *B. cepacia* is one of the four notorious aerobic lactose nonfermenting, catalase-positive, oxidase-positive Gram-negative bacilli. Here, we presented a case of *B. cepacia* bacteremia first of its kind, which is multidrug resistant (MDR) in a young immunocompetent patient having dengue and scrub typhus coinfection and reviewed the literature of *B. cepacia* infection in immunocompetent adult patients.

**CASE REPORT**

A 25-year-old male with no previous comorbidities was admitted in a hospital with a history of high-grade fever for 6 days, diffuse abdominal pain for 2 days, and one episode of hematemesis. Laboratory workup revealed anemia, thrombocytopenia, hyperbilirubinemia with raised transaminases, normal coagulation profile and renal function (hemoglobin 10 g/dl, total leukocyte count 15,000/cumm, serum creatinine 1.0 mg/dl, bilirubin (total/direct) 1.9/1.0 mg/dl, SGOT/SGPT/ALP 3390/1150/93 IU/L, INR 1.0) dengue NS1 antigen positive, malaria antigen and smear negative, chest radiograph, and computed tomography (CT) scan [Figure 1a] showed dense bilateral lower lobe consolidation. In the next 2 days, he developed...
acute respiratory distress syndrome (ARDS) requiring intubation and mechanical ventilation and septic shock, for which broad-spectrum antimicrobials (meropenem and doxycycline) were started, and he was referred to Intensive Care Unit (ICU) of our institute for further management. At the time of admission in ICU, he had fever (core temp 40°C), heart rate of 130/min, blood pressure (invasive) 130/70 mmHg on norepinephrine infusion (0.1 µg/kg/min), sedated with midazolam (3 mg/h) and fentanyl (100 µg/h), and on mechanical ventilation (PC/PEEP: 20/10 cm H₂O, respiratory rate 30/min, FiO₂:0.8). ICU severity scores were as follows: APACHE II 20 and SOFA 12. Initial laboratory workup at admission showed improving thrombocytoopenia and liver function tests. Workup for tropical infections revealed positive dengue IgM and positive scrub typhus serology. Procalcitonin was elevated at 6.5 ng/ml. Due to severe ARDS (PaO₂/FiO₂ ratio 100), he was proned for 12 h, after which his oxygenation status improved (PaO₂/FiO₂ ratio 220). Broad-spectrum antimicrobials were continued. However, he had persistent high-grade fever (core temperature 40°C) for the next 3 days despite recovering from septic shock. Three blood cultures drawn at admission grew *B. cepacia* which was resistant to amikacin (MIC >32), aztreonam (MIC >16), ceftazidime (MIC >16), and cefoperazone–sulbactam (MIC 16:64) and sensitive to levofloxacin (MIC: 2), meropenem (MIC: 2), and trimethoprim–sulfamethoxazole (MIC <0.5/9). Furthermore, endotracheal aspirates at ICU admission revealed the growth of methicillin-resistant *Staphylococcus aureus* (sensitive to linezolid and vancomycin). Antimicrobials were changed to doripenem, linezolid, and cotrimoxazole. Over 24–36 h following change of antimicrobials, fever reduced in intensity, which was followed by successful extubation 48 h later. Repeat blood cultures after 1 week of therapy with cotrimoxazole were sterile. Procalcitonin decreased to 0.5 ng/ml. CT imaging at discharge showed cavitory lesions with bronchiectatic changes in the lungs [Figure 1b] which resolved after few days.

**DISCUSSION**

This is the first reported case of MDR *B. cepacia* bacteremia in an immunocompetent host. The patient might have acquired infection during his earlier hospital stay. We verified that there was no epidemic of such infections in both the hospitals.

In the largest series (95 patients) of *B. cepacia* bacteremia from Taiwan, Liao *et al.* reported that 96% patients had more than one underlying disease. After personal communication with the corresponding author, we did not receive any information as to whether there was any immunocompetent patient in their cohort.[5]

In literature review, we found only nine case reports of *B. cepacia* in immunocompetent adult patients (age 25–66 years; male/female 7/1) as cited in Table 1. Sites of infection predominantly were lungs in five cases,[3,5,7,9] with one each in synovial fluid,[4] urachal abscess,[6] and one in blood causing native valve endocarditis.[8] In our case, it was in bloodstream infection.

*B. cepacia*, also labeled as “*B. cepacia* complex (BCC)” or nonmeliod *Burkholderia* species,[11] was previously mislabeled as *Pseudomonas cepacia* before the availability of automated technologies such as matrix-assisted laser desorption ionization, and time-of-flight mass spectrometry/Phoenix/Vitek-2/WalkAway systems. This is of great concern to the clinician as therapy differs for these organisms, which may lead to poor outcome. BCC is intrinsically resistant to first- and second-generation cephalosporins, aminoglycosides, antipseudomonal

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**Table 1: Burkholderia cepacia infections in immunocompetent adult patients**

| Author, year (reference) | Age/sex | Diagnosis | Site of infection | Treatment | Outcome |
|--------------------------|---------|-----------|-----------------|-----------|---------|
| Cabrera and drake 1975[5] | 66/male | Acute febrile illness | Lungs | Cefalothin and Kanamycin | Survived |
| Kothari *et al.*, 1977[4] | 58/female 32/male | Septic arthritis, CAP | Synovial fluid, Lungs Urachal abscess | Gentamycin, Quinolone Amoxicillin/clavulanic acid | Survived, Survived |
| Waterer *et al.*, 1999[5] | 56/male | Acute omphalitis with urachal abscess | Lungs Urachal abscess | | Survived, Survived |
| Hsu *et al.*, 2005[6] | 60/male 66/male 77/female | CAP, Spinal compression fracture with stroke and mitral stenosis (native valve endocarditis) | Lungs Blood | Ciprofloxacin, Levofloxacin Ceftazidime | Survived, Survived |
| Bayram *et al.*, 2011[7] | 32/male 25/male | CAP, CAP with pyopneumothorax | Lungs Blood | Ceftazidime Doripenem and cotrimoxazole | Survived, Survived |
| Bayram *et al.*, 2011[7] | 32/male | CAP, CAP with pyopneumothorax | Lungs Blood | Ceftazidime | Survived |
| Ki *et al.*, 2011[7] | 32/male | CAP, CAP with pyopneumothorax | Lungs Blood | Ceftazidime | Survived |
| Karanth *et al.*, 2012[8] | 32/male 25/male | CAP, CAP with pyopneumothorax | Lungs Blood | Ceftazidime Doripenem and cotrimoxazole | Survived, Survived |

CAP: Community-acquired pneumonia
penicillins, and polymyxins due to altered outer membrane permeability barrier (OMP) and efflux pumps.\[^{10}\] Acquired resistance is through production of beta-lactamases and modifying enzymes other than beta-lactamases. Usual treatment regimens include ceftazidime, minocycline, meropenem, or cotrimoxazole. Majority of BCC are susceptible to ceftazidime,\[^{2}\] but the BCC isolated in our case was resistant to this agent with high MIC (>16) which required administration of cotrimoxazole. Our case highlights that \textit{B. cepacia} may complicate community-acquired infections even in immunocompetent patients.

**CONCLUSION**

\textit{B. cepacia}, a microorganism causing infections in immunocompromised hosts, can complicate community-acquired infections as seen in our patient who is immunocompetent host. This organism historically described with low virulence can cause life-threatening infections. Proper identification of this organism and antibiotic susceptibility testing with the help of available automated technologies along with timely treatment with antimicrobials forms the key to management, due to varied resistance patterns noted.

**Consent**

Informed consent was obtained. All patient information has been de-identified to protect patient privacy.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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