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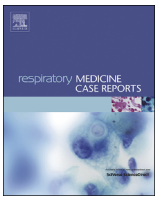

Case Report

Hospital-acquired pneumonia due to Burkholderia cepacia in a thalassemia pregnancy with postoperative eclampsia: A case report

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ABSTRACT

There are limited case reports on individuals infected with *Burkholderia cepacia* who do not have typical risk factors, particularly pregnant women with beta-thalassemia. A 34-year-old pregnant female with beta-thalassemia trait and hypertension was admitted to the hospital. The patient was diagnosed with eclampsia and underwent a cesarean section. After two days following the surgery, the patient experienced hospitality-acquired pneumonia. *B.cepacia* was isolated from blood cultures, and antibiotic susceptibility testing indicated sensitivity to trimethoprim/sulfamethoxazole and levofloxacin. The patient responded to antibiotic treatment. These findings highlight the importance of prompt diagnosis and appropriate treatment in managing *B.cepacia* infections in pregnant beta-thalassemia patients.

1. Introduction

Burkholderia cepacia complex (BCC) is a group of gram-negative bacteria [1]. Members of the BCC group are commonly found in environmental habitats such as soil, water, and various sources within hospitals [2]. To date, at least 20 closely related species within the BCC group have been identified [1]. BCC can cause opportunistic infections with a wide range of clinical symptoms, ranging from asymptomatic carriage to life-threatening conditions such as sepsis and pneumonia [3,4]. The most recognized risk factors in patients with BCC infection include cystic fibrosis, chronic granulomatous disease, and immunosuppression [4–6]. There are limited reports of BCC as a disease-causative agent in pregnant women [7–9] and patients with thalassemia [10]. This is the first case report of sepsis caused by *Burkholderia cepacia* in a pregnant patient with beta-thalassemia undergoing a cesarean section due to eclampsia in Vietnam.

2. Case presentation

A 34-year-old female patient at 36 weeks of pregnancy was admitted to Obstetrics Hospital due to uncontrolled hypertension, with a blood pressure of 180/100 mmHg. The patient has a history of beta-thalassemia trait characterized by mild anemia. There is no previous diagnosis of diabetes, cancer, alcohol addiction, human immunodeficiency virus infection, use of immunosuppressive medications (including corticosteroids), or other serious conditions. The fetus was developing normally during the pregnancy, although the patient experienced hypertension without treatment. At Obstetrics Hospital, the patient was diagnosed with eclampsia, leading to the

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decision to perform a cesarean section. Additionally, a urinary catheter was inserted to monitor urine output. On the second day following the surgical operation, the patient was presented with a high-grade fever (39 °C), rapidly progressing respiratory failure, productive cough, and pleuritic chest pain. Laboratory tests revealed a white blood cell count (WBC) of 18.6 k/uL with a neutrophil percentage of 92.7%. The C-reactive protein (CRP) level was measured at 191 mg/L, and the procalcitonin level was 2.23 ng/mL. A chest X-ray showed diffuse bilateral infiltrations involving both lung fields (Fig. 1A). The patient's condition rapidly deteriorated, requiring oxygen therapy via an oxygen mask of 10 L per minute to maintain a peripheral oxygen saturation (SpO_2) of 92% (with a PaO_2/FiO_2 ratio of 75). Afterward, the patient was promptly transferred to Cho Ray Hospital. Upon admission, the patient had a blood pressure of 100/60 mmHg, a heart rate of 90 beats per minute, and a body temperature of 38 °C. Clinical examination revealed tachypnea, accessory respiratory muscle involvement, and fine crackles auscultated bilaterally in the lungs. The surgical incision site showed no purulent discharge. Intravenous antibiotic therapy included imipenem/cilastatin 500 mg/500 mg four times daily, levofloxacin 750 mg once daily, and vancomycin 1000 mg twice daily, following the hospital's treatment protocol for hospital-acquired pneumonia.

The patient underwent bacterial culturing on blood, sputum, and urine samples. The culture results showed negative findings for sputum and urine samples. However, blood cultures at two separate sites concurrently revealed growth of *B.cepacia*, identified using the VITEK MS (Biomerieux, France) system through matrix-assisted laser desorption ionization time-of-flight (MALDI-TOF) methodology. Utilizing the automated devices based on microdilution susceptibility testing of the VITEK MS system, antibiotic susceptibility testing indicated that *B.cepacia* was sensitive to trimethoprim/sulfamethoxazole and levofloxacin while resistant to ticarcillin/clavulanic acid.

After seven days of antibiotic treatment, the patient's oxygen requirement decreased from 10 L per minute with an oxygen mask to maintaining a SpO_2 level above 94% without supplemental oxygen. A second chest X-ray revealed significant improvement in the infiltrative lung lesions in both lung fields (Fig. 1B). Procalcitonin levels decreased from 22.3 ng/ml to 0.64 ng/ml, CRP levels decreased from 190.9 mg/L to 33.4 mg/L, and WBC counts decreased from 18.6 G/L to 13.32 G/L. The patient was discharged with a 7-day prescription of levofloxacin 750 mg orally once daily and trimethoprim/sulfamethoxazole 960 mg orally twice daily.

After seven days of discharge, the patient had a follow-up examination, which revealed a complete resolution of respiratory symptoms. A chest X-ray revealed the complete absence of any residual abnormalities (Fig. 2). The WBC counts and CRP levels returned to normal baseline. Following the completion of a 14-day course of oral antibiotic therapy with levofloxacin and trimethoprim/sulfamethoxazole, the patient showed no signs of recurrent infection during the 3-month follow-up period.

3. Discussion

Our case report describes an unusual manifestation, including sepsis and hospital-acquired pneumonia, caused by *B.cepacia* in a pregnant patient with thalassemia who underwent a cesarean section due to eclampsia. Prompt administration of antibiotic treatment contributed to successfully managing this case report.

BCC is primarily recognized for the ability to cause infections in individuals with cystic fibrosis [1,4,11], chronic granulomatous disease [11], and compromised immune systems [1]. There are increasing reports of BCC infections among patients without cystic fi-

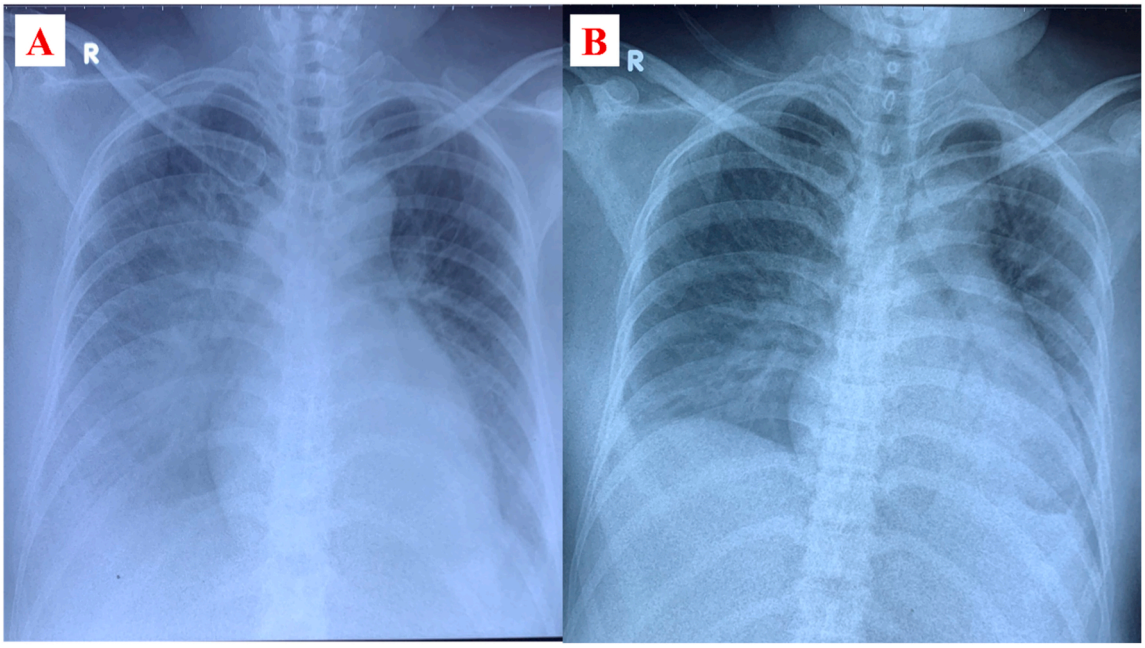

Fig. 1. Chest X-rays on day 1 (Fig. 1A) and day 7 (Fig. 1B) of the disease. Fig. 1A shows significant consolidation and bilateral infiltrates in both lung fields. Fig. 1B shows a remarkable reduction in lung parenchymal infiltrations.

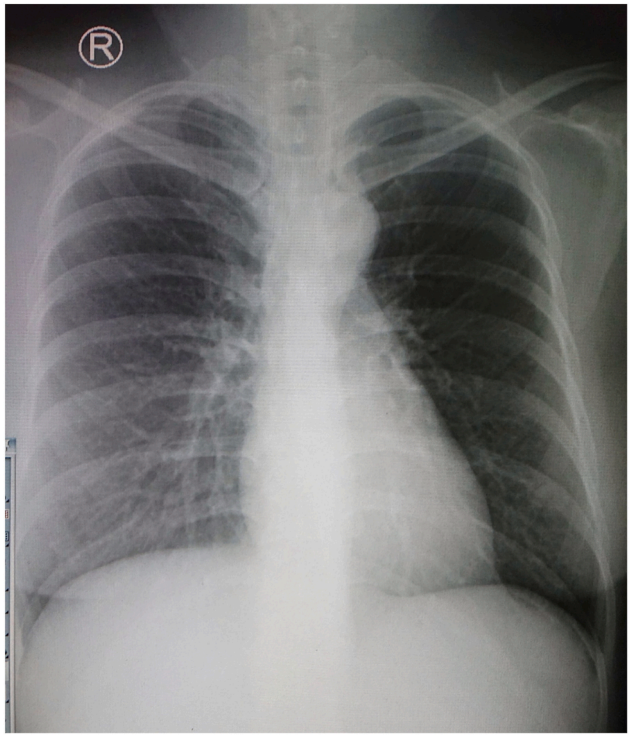

Fig. 2. Chest X-ray performed during the follow-up visit (on day 14 of the disease) revealed the absence of any residual lung parenchymal infiltrations.

brosis [12,13] or presenting with clinical symptoms beyond the respiratory system [14]. However, characteristics such as pregnancy, thalassemia, and eclampsia have not been reported in association with BCC infections. There are limited reports on cases of BCC infection in pregnant patients without cystic fibrosis [7,9]. Ishtiaq reported a case of a 34-year-old pregnant patient at seven months of pregnancy who experienced eclampsia and sepsis caused by ceftazidime-resistant *B.cepacia*, and ultimately resulted in death [9]. Stavroula reported a case of *B.cepacia* colonization in the vagina of a pregnant patient, which subsequently led to cervical insufficiency, preterm birth, and neonatal mortality [7]. Pregnancy has been shown to reveal reductions in the levels of T cells, natural killer cells, phagocytosis, and Th1 responses. The most pronounced changes are observed during the third trimester, which may increase the risk and severity of specific infections [8]. Furthermore, hemoglobinopathies such as sickle cell disease [12] and thalassemia [5] have also been suspected to be a risk factor for BCC infections. Beta-thalassemia increases hemin levels, resulting in elevated heme oxygenase 1 production and decreased production of interferon-gamma and interleukin-10. Ultimately, this immune dysregulation can lead to immunosuppression and an increased risk of infection in patients with beta-thalassemia [15]. A retrospective study on 211 non-transfusion-dependent thalassemia patients reported a severe infection rate of 5.2%, with commonly encountered bacteria, including *Klebsiella* species and *Burkholderia pseudomallei* [10]. However, there are currently limited case reports regarding BCC infections in patients with thalassemia or pregnancy.

Our case report represents the unusual postpartum sepsis caused by *B.cepacia* in a pregnant patient with beta-thalassemia who underwent a cesarean section due to eclampsia. The patient did not have known risk factors for BCC infection, such as cystic fibrosis, chronic granulomatous disease, or immunocompromised status [2]. However, due to the limitations of the clinical case report, we cannot draw any conclusions regarding the association between pregnancy, thalassemia, and BCC infection. Several other risk factors have been reported, such as renal failure requiring dialysis [13], abdominal surgery [13], the presence of a central line [13], and cancer [14]. Furthermore, in this case, the possibility of BCC colonization in the vaginal tract [7] or transmission of BCC through medical devices [2] such as disinfectants or urinary catheters cannot be ruled out. Therefore, the hypothesis concerning whether pregnancy or beta-thalassemia increases the risk of *B.cepacia* infection needs to be validated by future studies.

Despite numerous studies on BCC infections in cystic fibrosis patients, there is still a lack of optimized treatment protocol for this population [4]. The evidence regarding treating BCC infection in non-cystic fibrosis patients is even scarcer. Overall, a recommended antibiotic treatment protocol typically spans from 10 to 21 days and involves the combination of at least two antibiotics that exhibit sensitivity based on the antibiotic susceptibility testing results [2,4,14]. BCC has shown high resistance to colistin, whereas trimethoprim/sulfamethoxazole is considered first-line therapy for treatment [2,4]. In cases where patients are intolerant or allergic to trimethoprim/sulfamethoxazole, alternative treatment options include ceftazidime, meropenem, and piperacillin [6]. Quinolone and minocycline have also shown efficacy against BCC [2,14]. However, treating BCC infections remains a significant challenge due to the potential for antibiotic resistance during treatment and the progression to chronic infection. In our case, the patient received an additional 14-day course of oral antibiotics after discharge, including levofloxacin and trimethoprim/sulfamethoxazole, with the hope of successfully eradicating *B.cepacia* and minimizing the risk of developing antibiotic resistance.

In summary, this case adds to the literature another instance of a pregnant patient with beta-thalassemia who developed sepsis and hospital-acquired pneumonia caused by *B.cepacia* after undergoing a cesarean section due to eclampsia. Although a clear association has not been reported between pregnancy and thalassemia increasing the risk of BCC infection, clinicians should be aware of the potential for BCC infection in these populations to ensure optimal management.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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We have not received substantial contributions from non-authors.

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