Disseminated Pyomyositis Due to *Burkholderia cepacia*: A Case Report

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Abstract

Pyomyositis is a tropical infection affecting skeletal muscles manifesting as high-grade fever with pain in the affected limbs usually caused by Gram-positive microorganisms. Gram-negative organisms causing pyomyositis is uncommon but has been reported. *Burkholderia cepacia* is a Gram-negative nonfermenter causing opportunistic infections in immunocompromised patients, has been reported to cause pyomyositis only once before. We report a case of *B. cepacia* pyomyositis in a patient with no history of immunocompromised status, manifesting as disseminated infection with hemophagocytic syndrome presenting to our intensive care unit.

Keywords: *Burkholderia cepacia*, disseminated infection, pyomyositis

Introduction

Tropical pyomyositis is an infection of large skeletal muscles caused by Gram-positive microorganisms like *Staphylococci* in >90% of cases.[1,2] In immunocompromised states, this infection has been reported with Gram-negative microorganisms such as *Hemophilus, Enterobacter, Klebsiella species, Pseudomonas* species, and others.[3] Pyomyositis due to *Burkholderia cepacia*, a Gram-negative nonfermenter has only been reported once before in literature in a patient of cystic fibrosis.[4] We report an otherwise healthy young adult with a recent diagnosis of diabetes mellitus, presenting with *B. cepacia* pyomyositis manifesting in its disseminated form. To the best of our knowledge, this is the first case in literature, of *B. cepacia* pyomyositis, presenting in disseminated form manifesting with severe sepsis and secondary hemophagocytic lymphohistiocytosis.

Case Report

A 35-year-old male, a school teacher by profession presented with chief complaints of fever, pain in the right thigh and right shoulder for the past 15 days. The pain progressed to the right foot, left foot and right index and left ring fingers of his hands over the next 4 days. He was prescribed oral amoxicillin-clavulanate and paracetamol locally, despite which fever and limb pains persisted. He was admitted to a private hospital for a week, where his laboratory investigations revealed neutrophilic leukocytosis and hyperglycemia. Ultrasonography of the right mid arm, right forearm, and right thigh showing echogenic intramuscular collections [Figure 1a-c] suggestive of pyomyositis and early stage of abscess formation. Aspiration of pus yielded Gram-negative microorganisms, but further identification could not be done at that center. He was prescribed intravenous antibiotics (piperacillin-tazobactam and vancomycin) and subcutaneous insulin therapy. As the clinical condition did not improve over the next 10 days, he was referred to our hospital. At admission, he was in respiratory distress and was referred to intensive care unit (ICU) after the initial diagnostics [Figure 2a and b]. On examination, he was in encephalopathy, septic shock with impending respiratory failure. Examination of both feet and hands revealed diffuse, warm, and tender swellings. Admission APACHE II and SOFA scores were 19 and 6, respectively. He was intubated (moderate acute respiratory distress syndrome: PaO_2/FiO_2: 160), ventilated with lung protective ventilation strategy along with initiation of vasopressor support

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(norepinephrine @ 0.1 mcg/kg/min) for shock. A repeat ultrasound at our center did not show any pus collections for aspiration; although, there were inflamed muscle membranes and fibrillary pattern of muscle was lost [Figure 1d]. Doppler screen of limbs did not reveal any venous thrombosis. A two-dimensional-echocardiogram did not reveal any features of infective endocarditis. Empirical antimicrobials (meropenem and vancomycin) were administered. The admission blood cultures and subsequent blood cultures were positive for *B. cepacia*. In view of pancytopenia (Hb 5.2 g/dl, total leukocyte count 2100 cells/cumm and platelet count of 40,000 cells/cumm), hyperferritinemia (serum ferritin 5500 ng/ml), and hypertriglyceridemia (serum triglycerides 300 mg/dl) alongwith hepatosplenomegaly, with a strong suspicion of secondary lymphohistiocytosis, a bone marrow examination was performed [Figure 3a] which showed features of hemophagocytosis alongwith growth of *B. cepacia* [Figure 3b] resistant to amikacin (minimum inhibitory concentration [MIC] >32), gentamicin (MIC >8), piperacillin tazobactam (MIC >32) with sensitivity to meropenem (MIC: 2), cotrimoxazole (MIC: <0.5/9), and levofloxacin (MIC: 2). There was no granuloma or necrosis in the aspirate and no growth of fungal elements. Once these culture reports were available, cotrimoxazole was added to his therapy. Shock and hypoxemia improved by day 5 of ICU stay. The patient was febrile (core temperature of 40°C) during the first 3 days of his stay, progressively improved and became afebrile by day 10 of ICU stay. Meropenem and vancomycin were stopped after 7 days, and cotrimoxazole was continued up to 14 days. After 15 days of ICU stay, he was transferred to ward from where he was discharged home.

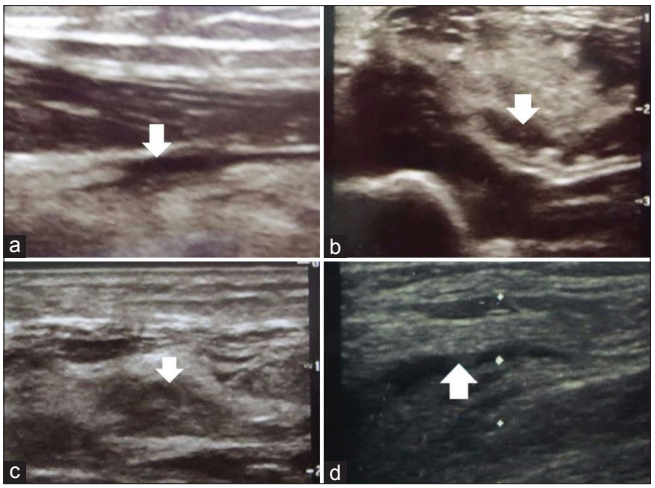

Figure 1: (a) Ultrasonography of thigh revealing a small pocket of hypo-echoic collection within the inter-fascial plane. (b and c) Ultrasonography of upper arm revealing bulky muscle which is heterogeneous in echotexture with loss of fibrillar pattern with few hypoechoic areas – collection pockets. (d) Ultrasonography of thigh done few days after intensive care unit admission suggestive of better delineation of muscle fibrillary pattern and relatively normal muscle bulk. Small pocket of collection is still there

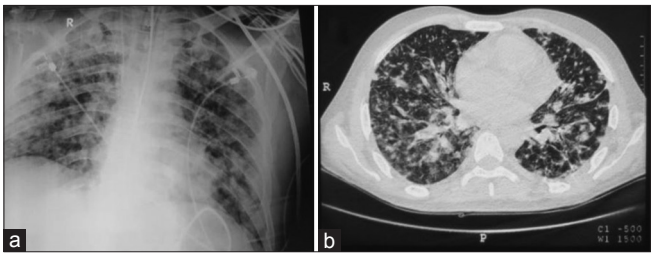

Figure 2: (a) Chest X-ray at admission to intensive care unit suggestive of bilateral diffuse alveolar infiltrates with well-preserved costo-phrenic angles and cardiac size. (b) Computed tomography chest at admission suggesting bilateral ground glass opacities with reticulation and small nodules distributed in both lung fields suggestive of acute respiratory distress syndrome

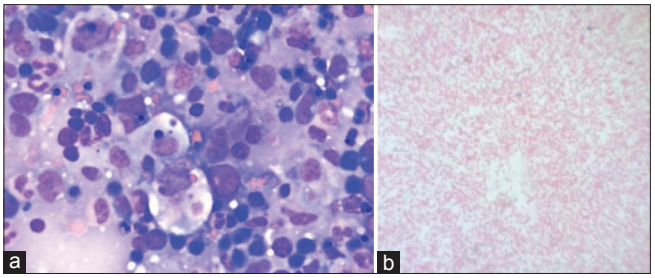

Figure 3: (a) Bone marrow smear showing increased histiocytes with prominent hemophagocytosis (Wright-Giemsa, ×400). (b) Motile Gram-negative straight bacilli after overnight incubation from the blood agar which are nonlactose fermenting catalase-positive and oxidase-positive

Discussion

Pyomyositis is an infection of tropics, involving skeletal muscles, caused most commonly by Gram-positive microorganisms like *Staphylococcus aureus*. Infection usually occurs in large striated muscles of the thigh, calf, gluteal region and shoulder regions causing exquisite pain and swelling.[1,2] The infection manifests in three stages, Stage 1: confined to muscles manifesting as fever with muscle pain and swelling, Stage 2: manifesting as suppuration or abscess formation in the involved muscles and Stage 3: disseminated form leading to bacteremia, infective endocarditis, pneumonia, septic arthritis, and rhabdomyolysis. Treatment of pyomyositis is with antibiotics and local drainage or debridement of the abscess.[1,2]

Gram-negative microbes causing pyomyositis have been reported to cause around 130 cases as reviewed by Gousseff *et al*. during the period 1979–2011.[3] *B. cepacia*, one of the species in so-called "*Burkholderia cepacia* complex" is a Gram-negative glucose nonfermenting rod historically described as having low virulence. Cepacia syndrome presents as fever, leukocytosis, pulmonary infiltrates, respiratory distress (necrotizing pneumonia), and septicemia in the form of infective endocarditis, septic arthritis, and abscesses (in implants and pacemaker pockets) predominantly in patients with immunocompromised states such as cystic fibrosis, chronic granulomatous disease, and diabetes mellitus and less often, in immunocompetent hosts.[4] Previously, this microbe was mislabeled as belonging to the pseudomonas group, but

currently, with automated diagnostic technologies, proper identification is possible. This is of importance, due to the varied intrinsic resistance profile of this organism.[5,6]

Our case is of importance because this is the first case of *B. cepacia* disseminated pyomyositis with the organism isolated from bone marrow and multiple blood cultures. The index patient was likely immunocompromised secondary to uncontrolled diabetes. MRI would have been useful in diagnosis but was not done due to multiple sites of involvement and initial critical status of the patient. Management through antibiotics is as effective as drainage techniques when definitive pus collection cannot be obtained.[7] Higher dosage of trimethoprim (15–20 mg/kg/day divided into three doses per day) should be considered, taking into account pharmacokinetics and dynamics in critically ill patients.[8,9]

Conclusion

Gram-negative organisms are increasingly being reported to cause tropical pyomyositis especially in immunocompromised patients. We report *B. cepacia* associated tropical pyomyositis and secondary lymphohistiocytosis in a young diabetic patient. *B. cepacia*, a microorganism with a varied intrinsic resistance profile, needs to be identified early by automated diagnostic technologies, to avoid inappropriate antimicrobial therapy.

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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Nil.

Conflicts of interest

There are no conflicts of interest.

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