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Case of Sepsis and Probable Septic Arthritis with *Plesiomonas Shigelloides* in a Patient with Sickle Cell Anemia

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Abstract

We present a case of sepsis with probable septic arthritis due to *Plesiomonas shigelloides* in a 10 year old female with hemoglobin SC disease. There have been three other cases reported in pediatric sickle cell disease (SCD) patients. Awareness among clinicians regarding this Gram negative organism is important as it can cause serious invasive infection in SCD patients, including sepsis and involvement of joints, lungs and intraabdominal organs. Clinical suspicion with early microbiologic diagnosis and timely use of appropriate antibiotics are key to the management of this infection and prevention of associated morbidity.

Keywords: Sickle cell anemia; Sepsis; *Plesiomonas shigelloides;* Septic arthritis; Gram negative

Background

Sickle cell disease (SCD) patients are at increased risk for Gram negative and unusual bacterial infections. *Plesiomonas shigelloides* is a Gram negative, facultative anaerobic rod, member of the family *Enterobacteriaceae* that has been isolated from environmental sources and a wide range of animals including mammals, birds, fish, water-dwelling reptiles and amphibians [1]. This report is the first to document a case of probable Plesiomonas septic arthritis in a pediatric SCD patient.

Case Report

A 10-year-old African American female known to have hemoglobin SC disease presented to the emergency department (ED) at Texas Children's Hospital in late September with one day of fever, vomiting and leg pain. She had eaten sushi five days earlier, but she had no other identifiable risk factors for infection. In the ED she was found to have tachycardia, tachypnea, hypotension (108/50 mm of Hg) and a temperature of 103.3°F. On physical examination, she demonstrated diffuse bilateral lower extremity pain; the remainder of the examination was normal. Laboratory evaluation included a mildly elevated white blood cell (WBC) count and low hemoglobin. Three sets of appropriate volume peripheral blood cultures (aerobic and anaerobic) were obtained. She received one dose of ceftriaxone in addition to fluid resuscitation (total 40 cc/kg of normal saline) and medications for pain control. Her chest radiograph was unremarkable. She was admitted to continue empiric therapy with cefotaxime for presumed sepsis.

Her fever initially resolved after the first dose of antibiotics, but her WBC count increased from 16,560 cells/UL on admission to 64,000 cells/UL and hemoglobin dropped from 11.6 gm/dl to 9.6 gm/dl on hospital day 2. The C-reactive protein (CRP) was elevated at 14.3 mg/dL and erythrocyte sedimentation rate (ESR) was 48 mm/hr. Admission blood cultures became positive within 48 hours for a non-lactose fermenting Gram-negative rod and her antimicrobial therapy was changed to ceftazidime to broaden coverage for possible pseudomonas infection. Subsequently, all six of the admission blood culture bottles obtained in the ED was positive for *Plesiomonas shigelloides*. The organism was susceptible to amoxicillin clavulanic acid, ceftriaxone, cefepime, levofloxacin, meropenem, piperacillin tazobactum and trimethoprim sulfamethoxazole. All blood cultures obtained after the initiation of antibiotics remained sterile. Infectious Diseases was consulted due to the unusual organism isolated in the blood and recurrence of fever after 48 hours (Figure 1).

On repeat examination on hospital day 2, she was found to have pain, swelling, warmth and limited range of motion of the right shoulder. Plain radiograph showed mild thickening and irregularity of the medial cortex of the right mid and distal humerus. Magnetic resonance imaging with contrast of the right shoulder showed moderate sized effusion with synovial enhancement and osteonecrosis of the right humeral diaphysis (Figure 2). Hence on day 3 of hospitalization, she underwent a right shoulder arthrotomy with exploration and drainage, and a drain was left in place which gave moderate sero-sanguinous discharge. Fluid from her shoulder joint was obtained, however no cell count was performed and cultures were sterile. The fever resolved after the 8th day of intravenous ceftazidime.

Upon clinical improvement and removal of the right shoulder joint drain, she was discharged after 11 days of hospitalization with instructions to continue treatment with amoxicillin-clavulanic acid to complete a total of 6 weeks of antimicrobial therapy for probable septic arthritis of the shoulder and possible osteomyelitis of the proximal humerus. At follow up after 4 weeks of therapy, she had remained afebrile and had improved range of motion of the right shoulder. A plain radiograph of her right shoulder was normal and her inflammatory markers had also normalized (CRP: 0.6 mg/dL; ESR: 17 mm/hr).

Discussion

P. shigelloides belongs to the family *Enterobacteriaceae* [1]. These microbes are Gram-negative anaerobic rods that have been associated with diarrhea and dysentery [2]. The primary natural reservoirs for *P. shigelloides* are water and soil surfaces as well as fish and other marine animals, especially oysters. The organism is recovered from freshwater and estuaries in temperate and tropical regions and occasionally from seawater during the summer months [3].

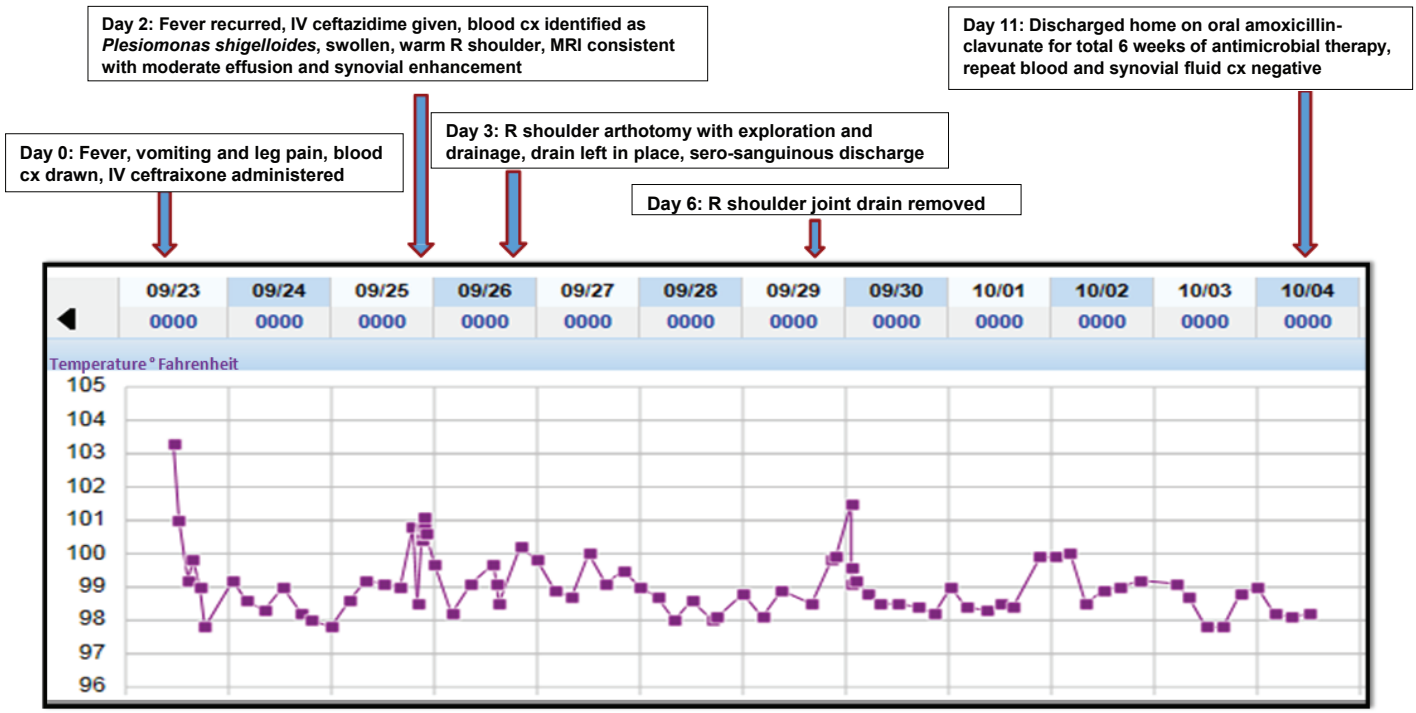

Figure 1: Clinical course of a sickle cell patient with Plesiomonas sepsis and probable septic arthritis, managed with intravenous (IV) ceftazidime and surgical drainage, showing clinical recovery
Abbreviation: Cx: culture; R: right

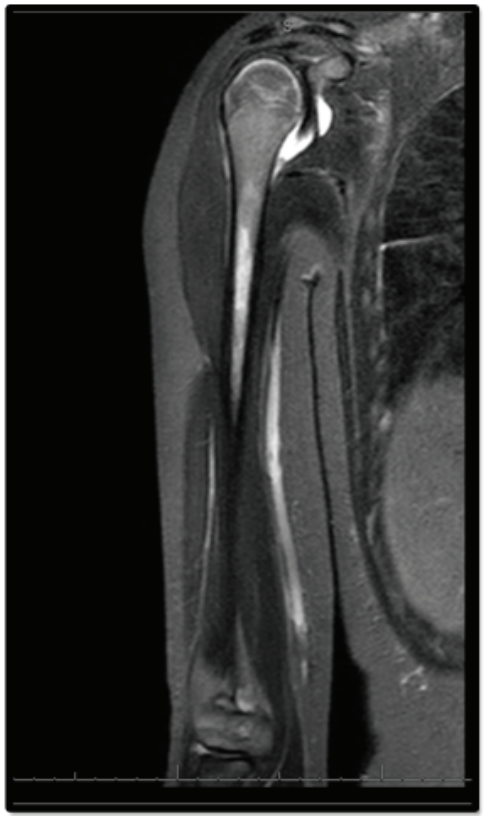

Figure 2: Magnetic resonance imaging with contrast of the right shoulder of a sickle cell disease patient with Plesiomonas sepsis showing moderate sized effusion with synovial enhancement and osteonecrosis of the right humeral diaphysis

Extra-intestinal *P. shigellosis* disease is less common than intestinal disease and it occurs in both immunocompromised and immunocompetent hosts [4,5]. Reported clinical manifestations include acute respiratory distress syndrome, disseminated intravascular coagulation, splenic abscess [6], ophthalmitis [7], neonatal meningitis [8,9], cellulitis [10,11] and orchi-epididymitis [12]. Though uncommon, *Plesiomonas* bacteremia has been associated with biliary tract disease, especially in older patients [13]. Invasive *P. shigelloides* infection has been associated with a mortality rate of 50% to 60% [14].

It is important to take notice of this organism in causing invasive disease in pediatric patients with sickle cell disease as it may lead to severe consequences. In addition to our case, three other cases of *P. shigelloides* sepsis have been reported in SCD patients (Table 1). Among them, two were associated with end organ disease (pneumonia and splenic abscess). There were no deaths reported. Similarly, our case illustrates the potential for *Plesiomonas* to cause serious infection in the context of bacteremia and sepsis in SCD patients and stresses the importance of having a high level of suspicion, and performing careful and repeated physical examinations to evaluate potential foci of infection such as a joint in patients with continuous or recurrent fever while on appropriate antimicrobial therapy. Development of joint pain in settings of high WBC count, MRI finding of moderate effusion, need for arthrotomy and drain placement are consistent with septic arthritis though synovial fluid culture remained negative. As osteonecrosis is a normal disease progression of underlying SCD, a joint infection could be missed. Moreover, the impaired reticulo-endothelial clearance associated with the disease has been shown to predispose patients with sickle cell anemia to bacterial infections [15].

P. shigelloides is inherently resistant to ampicillin, but amoxicillin or piperacillin in combination with beta-lactamase inhibitors (clavulanate and tazobactam) can be used as antiinfective agents. Isolates generally are susceptible to cephalosporins, quinolones and carbapenems [9,16-18].

Underlying disorder	Year	Age in years/ sex	Location	Presentation	Diagnosis	Susceptibility pattern	Complications	Antibiotics	Discharge home
Sickle cell disease*	1984	10/F	Los Angeles, US	Sepsis and pneumonia with effusion	Blood culture	Susceptible to moxalactam, gentamicin, chloramphenicol	Thoracentesis	Ampicillin, gentamicin, chloramphenicol, cefazolin, moxalactam	Day 14
Sickle Cell disease**	2001	13/F	New York, US	Sepsis and splenic abscess	Blood culture grew Gram-negative rods in 5 hours; identified on day 2	Not mentioned	ICU stays ~ 3 weeks; required intubation, chest tubes and exploratory laparotomy. Drainage of splenic abscess (900cc).	Cefotaxime, gentamicin; followed by ciprofloxacin orally for 14 days	Day 57
Sickle beta thalassemia zero***	2010	16/F	Sao Paulo, Brazil	Septic shock	Blood culture positive and organism identified on day 2	Resistant to ampicillin Susceptible to amikacin, gentamicin, trimethoprim-sulfamethoxazole, imipenem, ciprofloxacin, ceftriaxone	ICU stay 14 days; required intubation, chest tubes, exploratory laparotomy. Superinfection with MRSA+ (vancomycin susceptible)	Ceftriaxone, clarithromycin, metronidazole; followed by imipenem for 14days	Day 24
Sickle cell SC disease	2014	10/F	Houston, US	Sepsis and septic arthritis	Blood culture positive and organism identified on day 2	Susceptible to amoxicillin clavulanic acid, ceftriaxone, cefepime, levofloxacin, meropenem, piperacillin tazobactum, trimethoprim sulfamethoxazole	No ICU stay. Required arthrotomy and drainage of the right shoulder joint, drain left in place for 3 days	Ceftriaxone, ceftazidime; followed by oral amoxicillin-clavulanic acid for 6 weeks of total antimicrobial therapy	Day 11

Table1: Reported cases of sickle cell pediatric patients with *Plesiomonas* infection
*Mcneely D, Percy I, Craft C, Cohen I (1984) Plesiomonas: biology of the organism and diseases in children. Pediatr Infect Dis J 3: 176-181 [19].
**Ampofo K, Graham P, Ratner A, Rajagopalan L, Della-Latta P, et al. (2001) *Plesiomonas shigelloides* sepsis and splenic abscess in an adolescent with sickle cell disease. Pediatr Infect Dis J 20:1178-1179 [20].
*** Martins MA, Fernando FB, Viana JM, Teixeira GC, Nicolini EA, et al. (2010) Septic shock in a patient with sickle beta-zero thalassemia. Heart Lung 39: 335-339 [21].
+MRSA, methicillin-resistant *Staphylococcus aureus*.

Aminoglycosides are generally ineffective. Duration of therapy depends on the type of infection. No definitive source of infection was identified in our patient.

Early diagnosis with blood cultures and timely use of appropriate antibiotics were key to the management of this infection and daily evaluation to identify additional foci of infection resulted in a prompt diagnosis of probable septic arthritis, which required surgical drainage in addition to prolonged antibiotic therapy to resolve.

Conclusion

Ours is the first case of *P. shigelloides* bacteremia and sepsis associated with probable joint infection in a pediatric SCD patient. An increased awareness among clinicians regarding this Gram negative organism is necessary as *Plesiomonas* has been associated with sepsis and bacteremia, and has the potential to involve other organs like joints, spleen and lungs in vulnerable populations such as patients with SCD.

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