Salmonella Osteomyelitis of the Hand in an Infant with Sickle Cell Disease

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Summary: Salmonella osteomyelitis involving the hand is a rare, but potentially morbid, complication of sickle cell disease in children. This entity can be difficult to distinguish from the more frequent presentation of dactylitis, but accurate diagnosis is critical to direct proper treatment. We report on a 15-month-old patient with sickle cell disease who ultimately developed osteomyelitis of 1 hand after an acute vasoocclusive episode caused 4 extremity dactylitis. The case description illustrates the diagnostic and treatment challenges. *(Plast Reconstr Surg Glob Open 2015;3:e298; doi: 10.1097/GOX.0000000000000267; Published online 29 January 2015.)*

Sickle cell crises are painful episodes of acute vasoocclusion and focal ischemia caused when inelastic, sickle-shaped erythrocytes lodge in the small vessels.[1] Involvement of the fingers and toes, termed dactylitis, manifests as acute pain, swelling, and erythema and is difficult to distinguish from infections of the soft tissue or bone.

Although the 2 entities bear remarkable similarities to one another, the management and outcomes are quite different. Dactylitis is typically self-limiting and requires only hydration and pain management, and operative intervention is rarely indicated. Occasionally, more serious sequelae, such as growth plate injury, can occur.[2] In contrast, soft-tissue infections with or without osteomyelitis typically require operative drainage and 6–8 weeks of antibiotics. Delay in diagnosis can lead to significant morbidity and compromise finger function and viability.[3]

In this report, we present a toddler with sickle cell disease who presented with acute vasoocclusive episode of all 4 extremities and was ultimately found to have metacarpal and phalangeal osteomyelitis in 1 finger. He suffered a recurrence despite medical and surgical treatment. The case highlights the diagnostic and therapeutic challenges associated with this entity.

CASE REPORT

A 15-month-old African-American boy with sickle cell disease presented to his hematologist with right hand swelling. For 5 days before presentation, the patient had symmetric swelling of the bilateral hands and feet. The swelling in his left hand and both feet improved over the course of 2 days, but his right hand and ring finger remained swollen and erythematous.

Upon presentation to his hematologist, the patient was admitted to our hospital for inpatient observation. On exam, the patient was febrile to 39.6°C and irritable, with marked swelling of the proximal right ring finger, as well as diffuse dorsal and volar hand swelling and erythema. He had a frank abscess overlying the volar fourth metacarpal phalangeal joint (Fig. 1). Radiographs of the right hand demonstrated soft-tissue swelling but no osseous abnormalities (Fig. 2).

A bedside incision released 5 mL of purulent drainage, the cultures of which were subsequently positive for *Salmonella* serogroup D, resistant to ampicillin. The antibiotic profile was adjusted accordingly by the primary service.
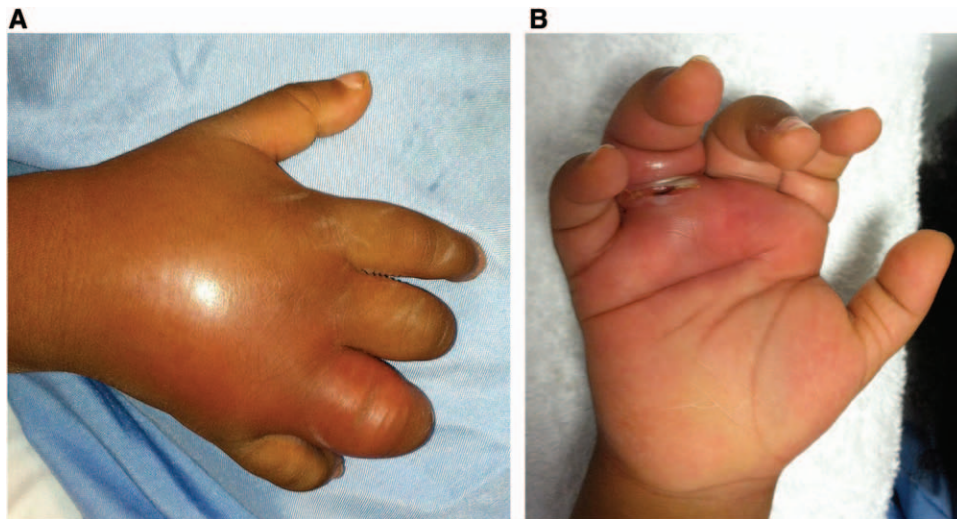
Fig. 1. Clinical appearance of right hand at time of initial consultation before incision and drainage. Swelling and erythema of proximal ring finger evident (A) and skin compromise over volar metacarpophalangeal joint crease (B).

Fig. 2. Initial posterior-anterior radiograph of the hand following incision and drainage and placement of penrose drain, without reactive changes to the metacarpal and phalanges of the ring finger.

Despite initial improvement in the right hand erythema and swelling, the patient remained febrile and his white blood cell count was persistently elevated at 20,000 cells/μL 24 hours after bedside drainage. The patient was taken to the operating room for exploration, at which time he was noted to have a collection of purulent fluid within the subperiosteal space of the right ring proximal phalanx. The adjacent volar, dorsal, and flexor compartments were unaffected. The patient subsequently defervesced and his white blood cell count normalized. His blood cultures remained negative, and his wound cultures from the operating room also grew *Salmonella* serogroup D, resistant to ampicillin. Infectious disease initiated a 6-week course of intravenous (IV) ceftriaxone for osteomyelitis.

Unfortunately, the patient returned approximately 4 weeks after completion of the 6-week course of IV antibiotics with a recurrent hand infection. Upon exploration in the operating room, he was found to have a 5 mL collection of purulent material dorsally, with severe osteomyelitic destruction of the ring finger metacarpal (Fig. 3). Neither the volar compartments nor the flexor tendon sheath were involved. Cultures were again positive for *Salmonella* serogroup D, resistant only to ampicillin. The infectious disease service initiated an additional 6-week course of IV ceftriaxone, followed by a 6-week course of oral therapy. The patient has been asymptomatic for 4 months following this regimen. Follow-up imaging demonstrated irregularity and osteosclerosis of the fourth metacarpal and proximal phalanx (Fig. 4).

Fig. 3. Posterior-anterior radiograph of the patient's hand following failure of initial surgical drainage and antibiotic course. Note the diffuse involvement of the ring finger metacarpal and proximal phalanx.

Fig. 4. Posterior-anterior radiograph obtained 3 months following presentation. Sclerosis but improved cortical irregularity and contour of ring finger metacarpal and proximal phalanx.

Statement of Ethics

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008 (5). Informed consent was obtained from all patients for being included in the study.

DISCUSSION

Acute sickle cell crises are marked by painful vasoocclusive episodes during which erythrocytes deform, limiting perfusion in the microcirculation of various organ systems including pulmonary, digestive and splenic, renal, central nervous system, and skeletal. In children, vasoocclusive crises affecting the bones are the most common acute symptom of sickle cell disease.[4] One possible explanation is that bone marrow in children is particularly hypercellular and susceptible to reduced blood flow and regional hypoxia and eventual infarction. Clinically, these episodes present with localized swelling, tenderness, and erythema. The most common sites for bony infarcts occur in the long bones where bone marrow is abundant, namely the femur, tibia, fibula, humerus, radius, and ulna.[4] In younger children, vasoocclusive episodes can involve the small bones of the hands and feet (dactylitis or "hand-foot syndrome"), which

still contain hemopoietically active bone marrow.[4,5] Occasionally, dactylitis is the first manifestation of symptomatic sickle cell disease.[6]

Although dactylitis is a far more common event than osteomyelitis in children with sickle cell disease,[7] the potential morbidity from a delay in treating osteomyelitis is grave.[8] Overall, *Staphylococcus aureus* is the most common cause of hematogenous osteomyelitis in children.[9] However, the ratio of *Salmonella* to *S. aureus* as the causative organism for osteomyelitis in the sickle cell population is 2.2:1.[10] It is believed that *Salmonella* bacteria enter the blood stream via microinfarctions of the gut as a result of impaired intestinal microcirculation. Bacteremia can then seed the infarcted bone, which serves as a nidus for fulminant infection. The long bones, specifically the femur, tibia, and humerus, are the most common sites of hematogenous osteomyelitis,[11] and infection of the small bones of the hand is a much more infrequent occurrence.[12,13]

Despite early identification and appropriate therapy, children with sickle cell disease and osteomyelitis involving the hand and wrist may have more complications than children without sickle cell disease. In their series comparing osteoarticular infections of the hand and wrist in children with and without sickle cell disease, Tordjman et al[12] demonstrated a 62.5% complication rate in patients with sickle cell disease, compared with 11.5%

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in patients without sickle cell disease. Additionally, the metacarpals and phalanges (88%) were most commonly affected in children with sickle cell disease, whereas distal radius and ulna (96%) were most frequently afflicted in children without sickle cell disease.[12]

Although our patient's initial symptoms were consistent with dactylitis, possibly triggered by dehydration from a recent gastrointestinal illness, a concurrent soft-tissue infection was readily diagnosed. The initial bedside drainage was performed because the radiographs were normal and the appearance of the infection was highly localized. Nevertheless, this approach proved inadequate. The persistence of symptoms and leukocytosis after the first debridement prompted further exploration in the operating room within 24 hours, and a subperiosteal collection of purulent fluid confirmed suppurative osteomyelitis of the metacarpal and phalanx. In retrospect, the duration of symptoms in our patient coupled with the possibility that the superficial abscess could originate from the underlying bone should have prompted more aggressive initial management of the hand infection in this patient with sickle cell disease.

CONCLUSIONS

Early and accurate identification of *Salmonella* osteomyelitis is critical to the successful management of this condition in patients with sickle cell disease. Empiric antibiotic therapy ensuring coverage of *Salmonella* is a key component in the initial treatment of patients with this presentation. Heightened awareness of the potential for a periosteal fluid collection in the small bones of the hand will guide optimal surgical intervention, thereby minimizing relapse and long-term morbidity associated with osteomyelitis.

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