The Journal of Foot & Ankle Surgery 52 (2013) 370–373

Contents lists available at ScienceDirect

The Journal of Foot & Ankle Surgery

journal homepage: www.jfas.org

Burkholderia pseudomallei Osteomyelitis of the Metatarsal in an Infant

Deeptiman James, MS [1], Vrisha Madhuri, MS, MCh [2], Abhay Deodas Gahukamble, MS [3], Lisa Choudhrie, MD [4], Padmaja Pancharatnam, MD [5]

[1] *Registrar, Paediatric Orthopaedic Unit, Christian Medical College, Vellore, South India*
[2] *Professor and Head, Paediatric Orthopaedic Unit, Christian Medical College, Vellore, South India*
[3] *Associate Professor, Paediatric Orthopaedic Unit, Christian Medical College, Vellore, South India*
[4] *Assistant Professor, Department of Pathology and Forensic Medicine, Christian Medical College, Vellore, South India*
[5] *Associate Professor, Department of Microbiology, Christian Medical College, Vellore, South India*

ARTICLE INFO

Level of Clinical Evidence: 4
Keywords:
antibiotic
bone
infection
melioidosis
saprophyte
surgery

ABSTRACT

Burkoholderia pseudomallei is an emerging cause of localized musculoskeletal infections. We report the case of a 9-month-old infant with isolated primary chronic osteomyelitis of the fifth metatarsal. Radiographs showed expansion and thickening of the cortex. The metatarsal had lytic lesions with scalloped margins; no periosteal reaction or sequestration was seen. Surgical debridement provided removal of infected material and adequate drainage by saucerization. *B. pseudomallei* was isolated from purulent material, and histologic examination revealed granulomatous inflammation. The child responded rapidly to a 2-week intravenous course of ceftazidime. The present case highlights the need for an awareness of melioidosis as a new differential diagnosis for a nontuberculous, granulomatous inflammation in those living in or visiting tropical regions.

Melioidosis, a saprophytic infectious disease, caused by gram-negative, soil-dwelling saprophyte *Burkholderia pseudomallei* is endemic in South East Asia and Northern Australia (1). It commonly presents as pneumonia and septicemia and is a very rare cause of osteomyelitis and septic arthritis (1,2). Disseminated septicemia can cause septic shock, which is fatal in nearly 80% of the cases (3). The presentation in children is usually community-acquired pneumonia, acute pharyngitis, parotitis, and septicemia, rarely infection is acquired through the birth canal (1,3,4). The present case report documents localized infection—a chronic osteomyelitis in an infant, highlighting the need for awareness of this entity as a differential diagnosis for persistent, recurrent, nonresponsive granulomatous infection in this age group. The etiologic diagnosis is easily missed, because the disease mimics other granulomatous inflammation processes. It requires prolonged treatment and can be fatal (1). Previous reports of orthopedic studies have mainly been of adults (5,6). Most have had an underlying systemic disease such as diabetes mellitus, thalassemia and chronic renal failure, or human immunodeficiency virus infection (1,5,7). Melioidosis as a possible differential diagnosis for a primary localized osteomyelitis in a child has not been previously highlighted.

Case Report

A 9-month-old infant presented with a swelling and discharging ulcers over the dorsolateral aspect of the left foot. The parents had noticed a swelling, that was insidious in onset, over the lateral aspect of the foot 1.5 months earlier. The patient had no history of trauma, fever, or swelling elsewhere in the body. No systemic manifestations were found. Primary treatment had been initiated at another institution, with a dorsal and plantar incision of the abscess and drainage of the purulent material. No culture was done at the initial treatment center. The child had developed persistent discharge from both incision sites, despite intravenous and oral antibiotics for more than 6 weeks. She presented to us with a 0.5-cm diameter ulcer over the dorsal lateral aspect of the left midfoot and a discharging sinus over the plantar aspect of the forefoot under the fifth metatarsal. She also had a subcutaneous abscess adjacent to the dorsal ulcer (Fig. 1). Both the dorsal ulcer and the plantar sinus were adherent to the fifth metatarsal, which was irregular, thickened, and tender. A culture at her initial presentation was taken from the superficial ulcer; however, it did not grow any organism. The radiographs revealed an expanded fifth metatarsal of the left foot with lytic areas at the proximal and distal ends of the shaft that were not associated with any periosteal reaction. Thickening of the metatarsal cortex was present, and the scalloped margins of the lytic lesions led us to suspect a granulomatous inflammation and investigate for an underlying pathologic entity (Fig. 2).

The laboratory investigations showed a C-reactive protein and total leukocyte count elevated to 64.1 mg/L and 14,200/mm^3,

Financial Disclosure: None reported.
Conflict of Interest: None reported.
Address correspondence to: Vrisha Madhuri, MS, MCh, Professor and Head, Paediatric Orthopaedic Unit, Christian Medical College, Vellore, India.
E-mail address: madhuriwalter@cmcvellore.ac.in (V. Madhuri).

1067-2516/$ - see front matter © 2013 by the American College of Foot and Ankle Surgeons. All rights reserved.
http://dx.doi.org/10.1053/j.jfas.2012.12.009

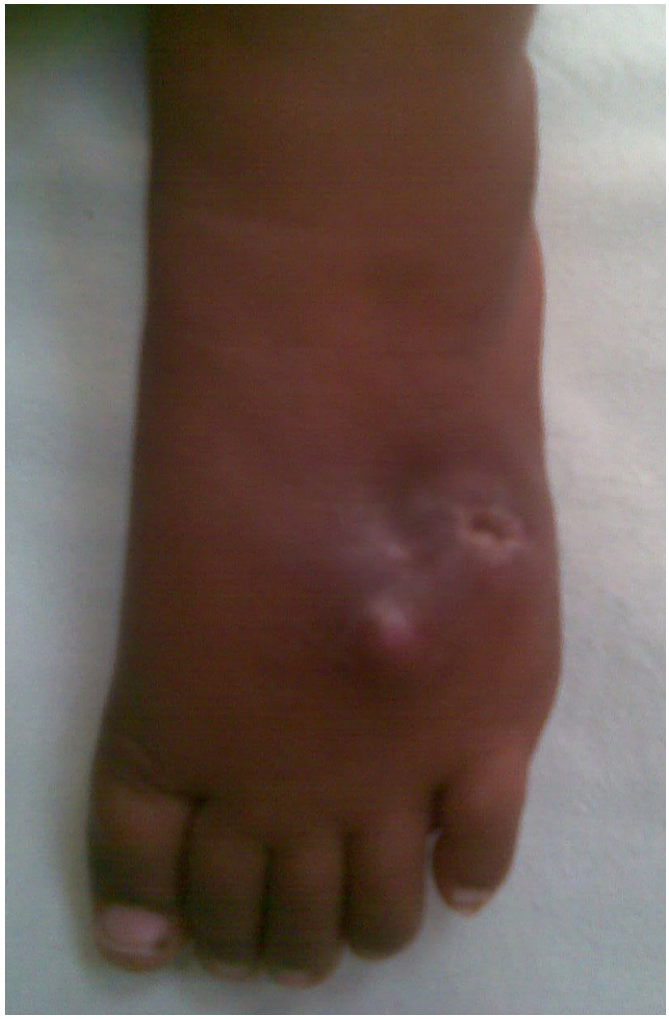

Fig. 1. Infant's left foot showing a puckered scar, a subcutaneous abscess on the dorsum, and an ulcer overlying the fifth metatarsal.

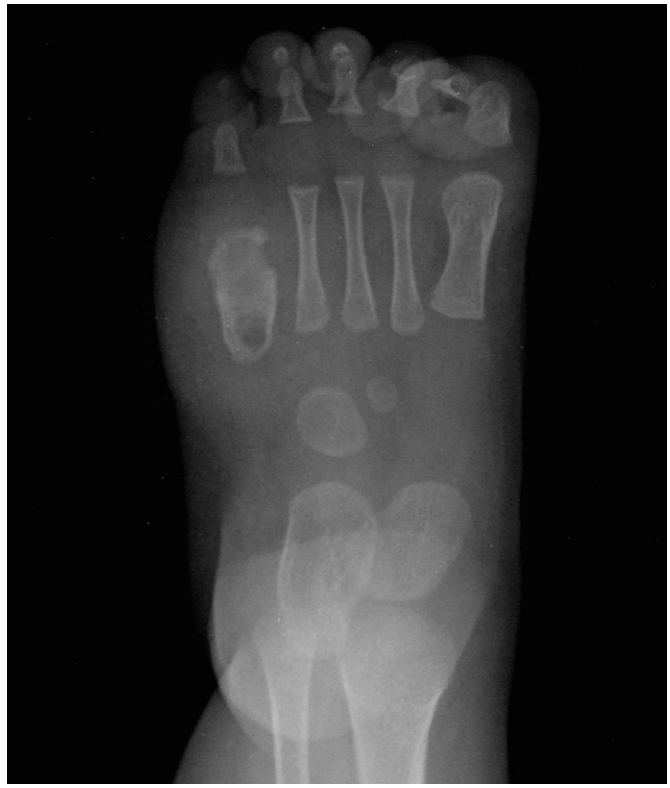

Fig. 2. Anteroposterior radiograph of the foot showing an expanded fifth metatarsal, scalloped outline, a cystic lesion in the proximal and distal part, and no sequestrum.

respectively. The bone scan showed uptake only in the fifth metatarsal. The investigations did not reveal an immunocompromised status. A culture from a throat swab did not show any growth. A differential diagnosis of tuberculosis and eosinophilic granuloma with a possible secondary infection was considered on the basis of the clinical features and findings from radiology studies and preliminary investigations.

Debridement of the fifth metatarsal lesion, including drainage of the subcutaneous abscesses, saucerization of the metatarsal, and curettage and excision of the sinus tracts, was performed through a dorsolateral incision. No bone matrix or filler was used. The wound was apposed with 2 sutures and allowed to close secondarily. The initial culture from the thick purulent material obtained showed nonfermenting gram-negative bacilli. Additional tests revealed *B. pseudomallei* was the organism isolated in the culture. The identification was based on the organism being a motile, nonfermenting, gram-negative bacillus that was oxidase positive and resistant to gentamicin and polymyxin B. Agglutination with *B. pseudomallei* antiserum was positive. Zeihl-Neelsen staining for acid fast bacilli was negative. No fungal elements were identified.

The histopathologic study of the specimen showed chronic osteomyelitis with granulomatous and pyogenic features (Fig. 3) and nonspecific chronic inflammation with pseudoepitheliomatous hyperplasia in the skin of the sinus tract. *B. pseudomallei* osteomyelitis affecting the metatarsal was diagnosed. The child was administered intravenous ceftazidime 75 mg/kg/d in 3 divided doses for 14 days. Rapid resolution of the lesion occurred, and it had healed within 1 week. The C-reactive protein level in the serum had decreased to $<$ 3 mg/L (normal for our laboratory $<$ 6 mg/L). The ceftazidime treatment was followed by a 3-month course of cotrimoxazole (trimethoprim-sulfamethoxazole 160/800 mg every 12 hours). The child had no subsequent recurrence during the next 2.5 years.

Discussion

Melioidosis has been increasingly diagnosed as a cause of pediatric infection in tropical countries, with reports mainly from Thailand, Vietnam, Northern Australia, Malaysia, and India (3,8). Fig. 4 summarizes the reports of melioidosis published in English-language studies and indicates the geographic distribution, acknowledging that the actual prevalence might differ owing to publication bias. Osteomyelitis and septic arthritis is reported in 1.5% to 8% of all cases, and very few reports of skeletal infections in children have been published (1,2). In the reported cases, septic arthritis involving the knee and ankle and osteomyelitis of the femur, tibia, and foot has been described in the adult, with the lower limb a preferred site owing to inoculation through the skin (5,7,9). *B. pseudomallei* osteomyelitis involving the foot bones is rarely, if ever, reported in children. *B. pseudomallei* is a saprophyte that survives in the clay layer, 25 to 30 cm underneath the soil surface and can be isolated from soil, stagnant streams, ponds, rice paddies, and even the market places in endemic regions (10). It enters the body through pre-existing skin lesions and causes localized musculoskeletal melioidosis (1). The