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CASE REPORTS

Pediatric blastomycotic osteomyelitis of the hand

Jason M. Erpelding · David W. Meister · Roger A. Daley

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Introduction

Accurate and timely diagnosis of uncommon blastomycotic hand infections can pose a diagnostic challenge. The clinical manifestations can be extremely variable by mimicking many common diseases or aggressive tumors. Blastomycotic hand infections are typically associated with disseminated infections and the patient must be treated with a high index of suspicion for additional sites of involvement [2].

Case Report

A 10-year-old, left-handed girl developed a non-productive cough shortly following a camping trip to a river bottom area of central Wisconsin that subsequently resolved within 2 weeks. She remained afebrile. Over the next few weeks, her left ring finger became mildly painful with slowly progressive swelling. There was no history of trauma to the finger. She presented to at an outside institution and was placed into a splint for a presumed benign bone tumor. She otherwise was a healthy, immunocompetent child. Except for the affected finger, there were no other areas of bodily discomfort. There was no cutaneous involvement at that time.

Her finger pain continued despite immobilization. Upon consultation in our clinic 1 month later, a 1×2 cm erythematous ulcerative lesion of the fourth web space had developed along the ulnar base of the proximal phalanx of the ring finger (Fig. 1). Circumferential swelling about the proximal phalanx and limited finger range of motion was noted.

J. M. Erpelding (✉) · D. W. Meister · R. A. Daley
Department of Orthopaedic Surgery,
Medical College of Wisconsin,
MCW Clinics at Froedtert East, 9200 W. Wisconsin Avenue,
Milwaukee, WI 53226, USA
e-mail: jerpelding@medicine.nodak.edu

Diagnostic investigation included complete blood count with a WBC of 13 K/μl, ESR 11 mm/h (normal 0–10 mm/h), CRP 0.6 mg/dl (normal 0–1.0 mg/dl), normal CMP and LDH. Radiographs of the ring finger revealed a large expansile mass of the ring finger proximal phalanx with periosteal reaction (Fig. 2). MRI of the finger revealed an expansile lesion of the proximal phalanx that was continuous with the ulcerating mass. The volar and ulnar cortices of the proximal phalanx were violated and there was involvement of the flexor tendon sheath and physis (Fig. 3a–c).

With MRI suggestive of tumor versus infection the patient was taken to the operating room for biopsy and cultures. Intraoperative preliminary frozen section was suggestive of infection. We then irrigated and debrided the wound bed, flexor tendon sheath, and curetted out the proximal phalanx (Fig. 4). Specimen microscopic examination and fungal stains showed broad based budding yeast and oral Itraconazole was initiated within 24 h. Cultures subsequently returned *Blastomyces dermatitidis* post-operative day 8. The patient had not received any antimicrobial or antifungal therapy prior to surgery. A computed tomogram

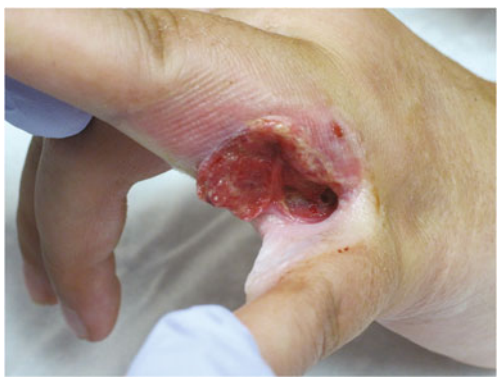

Fig. 1 Photograph of fourth web space ulcerative lesion with communicating sinus tract

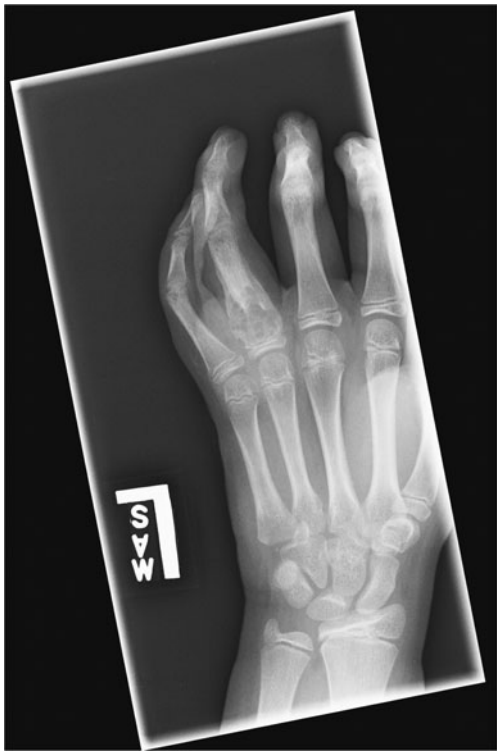

Fig. 2 Posteroanterior hand X-ray reveals expansile, erosive lesion of ring finger proximal phalanx with periosteal reaction

of the head was negative for any foci of infection. Chest radiograph revealed a small right upper lobe consolidation (Fig. 5). Bone scan incidentally showed increased uptake of the left proximal humerus. A lucent 1.5 cm focus within the left proximal humeral metaphysis adjacent to the physis was identified on standard shoulder radiographs (Fig. 6). The patient was asymptomatic at the shoulder.

The wounds, including the proximal phalanx, and flexor tendon sheath were irrigated and debrided again 2 days later with wound closure. Follow-up at 6 months showed complete healing of the lesion and full finger range of motion and sensation (Fig. 7a–b). Radiographs showed near complete remodeling of the phalanx with preservation of the physis (Fig. 8a–b). Itraconazole therapy was continued for 12 months.

Discussion

B. dermatitidis is a dimorphic fungus that can cause infections in immunocompetent individuals. The endemic region is the Midwestern, North Central, and Southeastern parts of

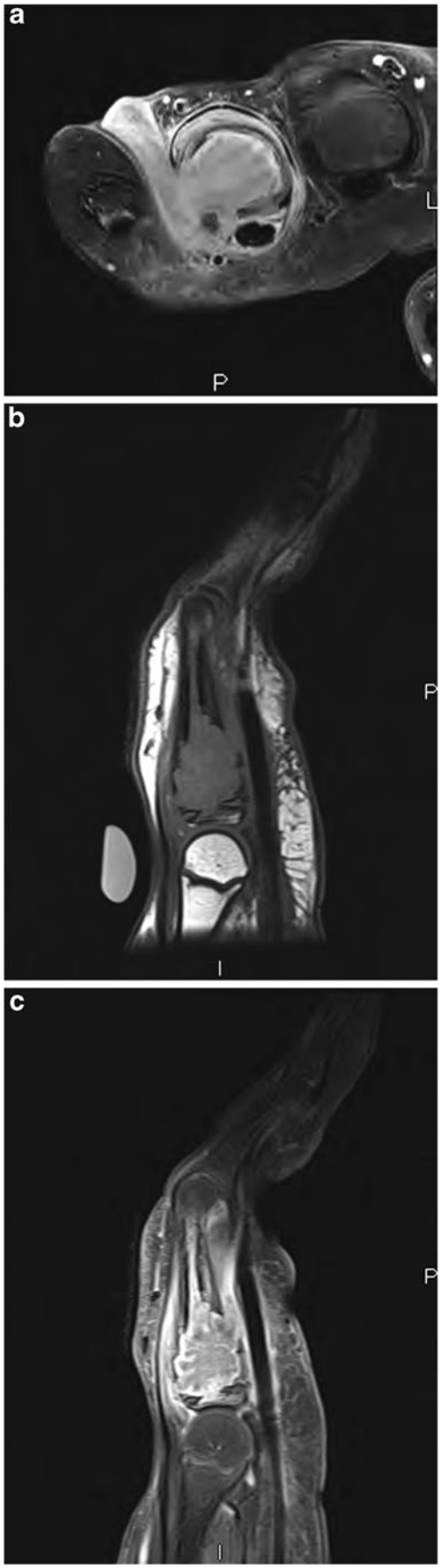

Fig. 3 **a** Axial T1 post-contrast fat-saturated MRI image demonstrating volar and ulnar cortical breaches and flexor tendon sheath involvement. **b** Sagittal T1 MRI image of ring finger. **c** Sagittal T1 post-contrast fat saturated MRI image of ring finger demonstrating involvement of the physis ▶

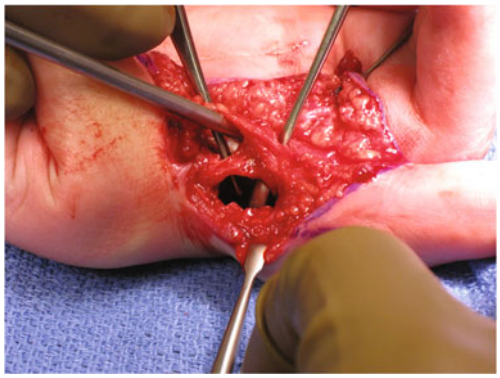

Fig. 4 Intra-operative photograph demonstrating erosion of volar and ulnar cortices of ring finger proximal phalanx with flexor tendon sheath involvement

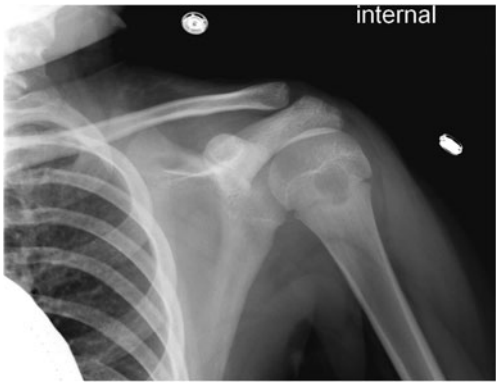

Fig. 6 Left shoulder anteroposterior internal rotation radiograph with radiolucent humeral metaphyseal lesion adjacent to physis

the United States, occurring at rates up to 8.3 per 100,000 people in these areas [9]. Approximately 3–11 % of cases occur in the pediatric population [13].

The most common mode of infection is through inhalation [12]. A primary pulmonary infection is often asymptomatic, and resolves quickly in the majority of cases [1]. If pulmonary macrophages are unable to neutralize the organism, it will develop into the yeast phase, multiply, and spread hematogenously after an incubation period of 3 weeks to 3 months [6, 8]. Infection may also occur from direct transcutaneous inoculation, such as an animal bite or exposure through breaks in skin to soil or tissue [2, 8]. Deep infections of the upper extremities will usually present as osteomyelitis, septic arthritis, or tenosynovial infections of the flexor or extensor compartments [9].

Patients with acute or chronic infection may complain of fevers, chills, night sweats, arthralgias, myalgias, weight loss, and pleuritic chest pain. The skin and musculoskeletal system are the primary sites of extrapulmonary disease. Cutaneous blastomycosis occurs in 80 % of all patients and frequently affects the dorsum of the hand [2, 9]. Skeletal disease occurs in 25–50 % of patients. These often present as osteolytic lesions. Osteomyelitis complicates systemic blastomycosis in 14–60 % of cases and is almost always associated with cutaneous and pulmonary involvement [8, 10].

Blastomycotic infections involving the hand are rare. Those affecting the pediatric hand are even less frequent. A recent 30-year series of pediatric and adolescent

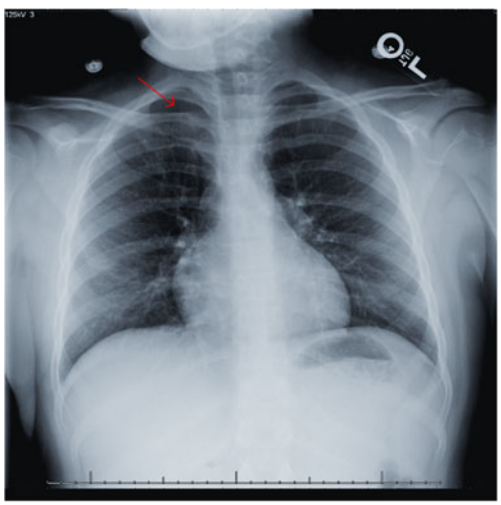

Fig. 5 Posteroanterior chest radiograph with subtle right upper lobe consolidation labeled with arrow

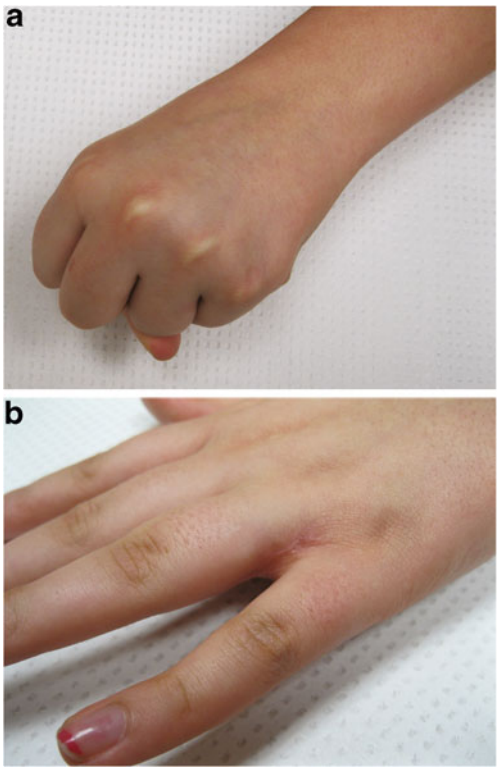

Fig. 7 **a** Photograph of dorsum of hand with full finger flexion. **b** Close-up view of healed fourth web space

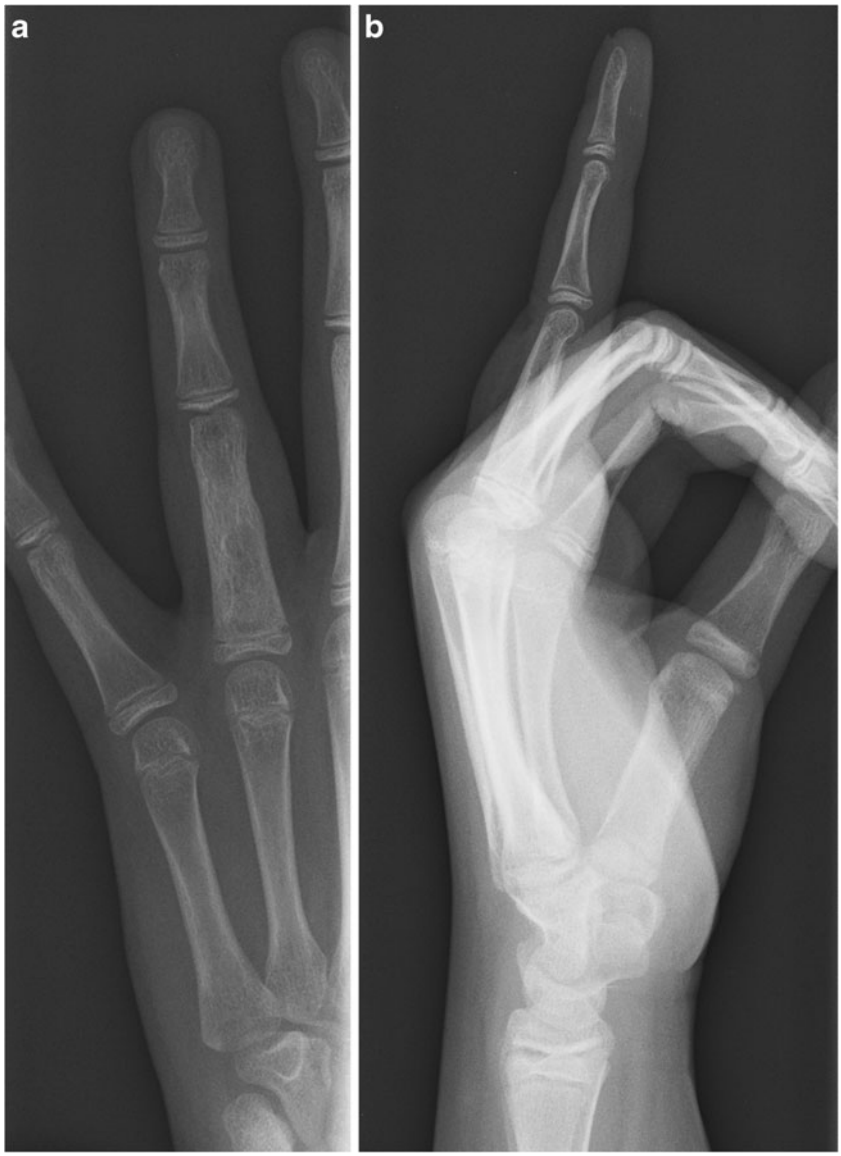

Fig. 8 **a** Anteroposterior 6 month follow-up radiograph of left fourth finger showing near complete proximal phalanx remodeling and preservation of the physis. **b** Lateral 6 month follow-up radiograph of left fourth finger

blastomycosis infections showed 21 % bone and joint of various sites of involvement, none of which involved the hand or wrist [6]. Oppenheimer et al. [11] had only four lesions involving the hands in their retrospective review of over a 40-year period. Bone is the third most common site of disease following lung and skin [12]. Approximately 25 % of cases with extra-pulmonary infection have osteomyelitis [3]. These infections typically progress in a rapid and aggressive fashion. This can occur more commonly from systemic dissemination or less commonly due to extension from a direct inoculation site of the hand [2]. Osteomyelitis can spread to adjacent joints, causing septic arthritis or involve soft tissues and form overlying subcutaneous abscesses, with draining sinuses progressing to ulcerating lesions. Radiographic evidence of blastomycotic osteomyelitis can range from discrete osteolytic lesions void of periosteal reaction to expansive destructive lesions with associated periosteal reaction [7]. Magnetic resonance imaging can be helpful with preoperative planning to ensure all involved structures are identified and thoroughly explored and debrided.

Culture using various media with chemiluminescent DNA probe verification is still the most sensitive method of diagnosis. If sufficient organisms are present, blastomycosis growth is typically present within 5–10 days; however, specimens with lower organism counts may require up to 30 days of incubation [12].

Timely and aggressive surgical debridement of abscesses and sinus tracts in conjunction with antifungal therapy are the mainstays of treatment. Immune status, clinical form and severity of disease dictate the appropriate treatment regimen. Amphotericin B is utilized in cases of moderately severe to severe infection particularly if life-threatening,

immunocompromised, pregnancy, or central nervous system (CNS) involvement [12]. Azole antifungal agents, most commonly Itraconazole as in this case, are utilized in moderate or milder infections and as step down therapy following 1–2 weeks of Amphotericin B treatment in patients without CNS involvement. Efficacy rates of Itraconazole monotherapy for moderate to mild disease involvement have been reported as 90–93 % in several studies [5]. Osteoarticular infections are typically treated for 12 months. Long-term suppressive Itraconazole therapy is often recommended in chronically immunosuppressed patients [4].

Blastomycotic hand infections in the pediatric population are exceedingly rare. A thorough history is necessary to reaching the diagnosis. As in this case, a subtle self-resolving cough shortly after an innocent camping trip were two, easily overlooked, key pieces of information that aided in directing the differential diagnosis. It is also of critical importance to assess patients for both pulmonary and extra-pulmonary sites of infection. In order to minimize loss of hand function a timely diagnosis and initiation of treatment are necessary. Particularly in endemic areas, a high index of suspicion for the possibility of a blastomycotic infection in all patients presenting with infectious or aggressive tumor-like lesions should be maintained.

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