Pediatr Radiol (2015) 45:1427–1430
DOI 10.1007/s00247-015-3299-7

CASE REPORT

Bone involvement by *Sporothrix schenckii* in an immunocompetent child

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Received: 12 May 2014 / Revised: 14 December 2014 / Accepted: 4 February 2015 / Published online: 17 February 2015

Abstract Sporotrichosis in children is rare, and its osteoarticular form is very unusual. Disseminated forms are described mostly in immunocompromised patients. We report a case of a 5-year-old immunocompetent boy with multiple suppurated cutaneous lesions that progressed to polyarthritis of the hands and feet. Radiographic imaging demonstrated multifocal lytic lesions. Sporotrichosis was diagnosed through biopsy and culture. This article describes the radiographic appearance of a rare manifestation of this disease. In areas of high prevalence, the diagnosis of sporotrichosis should be taken into account, even in immunocompetent patients, when dactylitis with lytic lesions is present.

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Keywords *Sporothrix schenckii* · Sporotrichosis · Bone · Immunocompetence · Radiography · Children

Introduction

Sporotrichosis is the most common subcutaneous mycosis in Latin America [1]. It is caused by the dimorphic fungus *Sporothrix schenckii,* and the most common form of infection is by the inoculation of the fungi through the skin and, rarely, by inhalation of conidia. The most classic presentation is associated with farming and other labor involving direct manipulation of the soil; however the zoonotic transmission by infected cats has been described as the main vector in Rio de Janeiro, Brazil [1]. After infection the clinical manifestations depend on a number of factors such as inoculum size, depth of inoculation and the immune status of the patient [1]. The extracutaneous form represents less than 1% of cases and is mostly related to some degree of immunosuppression caused by alcohol use or acquired immune deficiency syndrome (AIDS) [2]. Although skin involvement is well documented, there are few reports of systemic involvement; radiologic bone alterations caused by this fungus are very rare.

Case report

We present a 5-year-old boy with a 2-month history of nodular erythematous skin lesions that were slightly painful and scattered on the face, gluteal region and upper and lower limbs, and progressed to spontaneous suppuration accompanied by intermittent fever. After a month, swelling started in the boy's fingers and feet, with no redness or heat, and he was referred to our hospital for investigation. The boy's parents

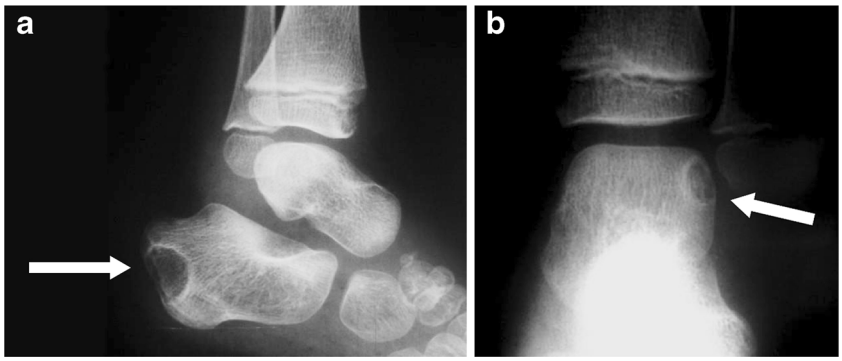

Fig. 1 Radiographs of the ankle in this case, a 5-year-old boy with osteoarticular sporotrichosis. **a** Lateral projection of the right ankle shows a well-defined lytic lesion with a sclerotic halo in the calcaneus (*arrow*). **b** Anteroposterior projection of the left ankle shows a well-defined lytic lesion with a sclerotic halo in the talus (*arrow*)

denied any history of disease and reported contact with cats. On physical examination the child was in very good general health. He had dactylitis in both hands and feet. Laboratory tests showed anemia and elevated erythrocyte sedimentation rate, C-reactive protein and lactic dehydrogenase. Human immunodeficiency virus (HIV) antibody test was negative, CD4/CD8 counts were normal, and the Venereal Disease Research Laboratory test was negative. Radiologic studies showed multifocal lytic lesions affecting the bones in both hands, feet and ankles and in the right elbow. Well-defined lytic lesions with sclerotic halo were identified in the talus and calcaneus and the distal metaphyses of the ulna, radius, humerus and tibia (Figs. 1 and 2). Permeative-pattern lytic lesions were predominant in the metadiaphyseal region of proximal and middle phalanges and the proximal phalanges of the feet, metacarpals and metatarsals; there was expansile and periosteal reaction associated with disruption of cortical bone (Figs. 3 and 4).

A hypothesis of fungal disease (sporotrichosis, paracoccidioidomycosis), tuberculosis or leukemia was considered. Biopsy and culture of skin lesions with secretion showed the growth of *Sporothrix schenckii*. Beginning the 7th day of hospitalization, the boy underwent treatment with amphotericin B for 14 days, with a subsequent change to itraconazole. The boy's clinical and radiologic findings improved; after 45 days of treatment radiographs showed bone sclerosis at sites of previous lytic lesions (Fig. 5).

Discussion

In Rio de Janeiro between 1997 and 2007, 67% of sporotrichosis cases occurred in women age 20–69, and 67% were caused by zoonotic transmission, 78% of these via cats [3]. The disease was rare in children, possibly because of its low exposure, with 6.6% of cases occurring in children younger than 12 years [3]. This differs from epidemiological data in Peru, where 60% of the cases occur in children [4].

Most cases are localized infections of the skin and subcutaneous tissue; these cases are not serious and respond well to antifungal agents [5]. The forms of sporotrichosis include the classic lymphocutaneous (most common), fixed cutaneous,

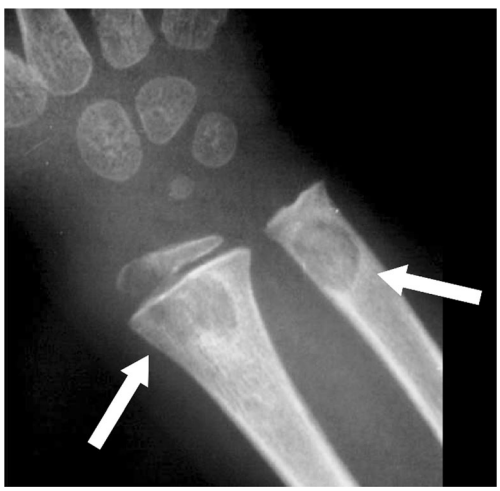

Fig. 2 Lytic lesions of the wrist. Anteroposterior radiograph of the left wrist shows well-defined lytic lesions (some with sclerotic halo) in the distal metaphyses of the ulna and radius (*arrows*)

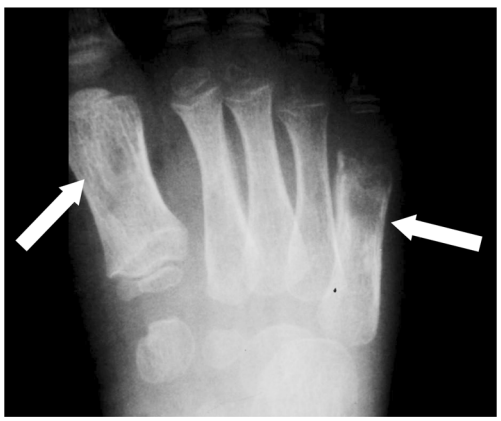

Fig. 3 Lytic lesions of the foot. Anteroposterior radiograph of the right foot shows permeative lytic lesions in metaphyseal metatarsal bones. Some lesions are expansile and associated with laminar periosteal reaction (*arrows*)

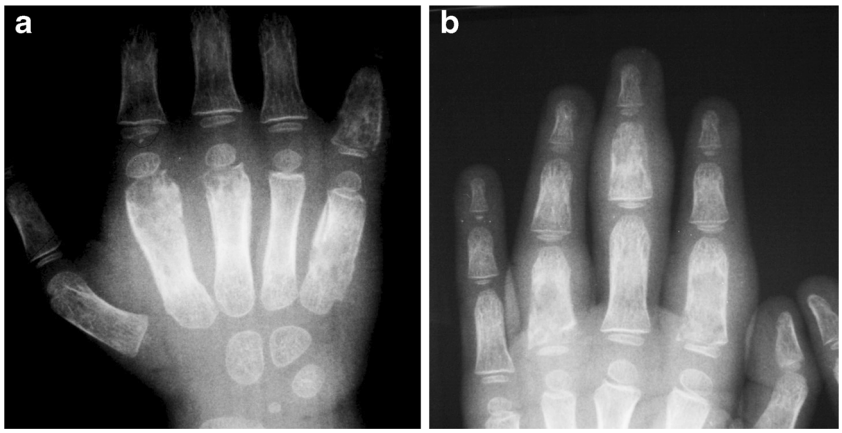

Fig. 4 Lytic lesions of the hands. Anteroposterior radiographs of the left hand (**a**) and right hand (**b**) show permeative lytic lesions at the metadiaphyseal region of metacarpal bones and phalanges, some of which are expansile and lead to cortical rupture and periosteal reaction, as well as subchondral osteopenia

disseminated cutaneous, primary pulmonary, and systemic. The disseminated form occurs in 5% of cases in Rio de Janeiro [3], and the hematogenous spread is rare in immunocompetent patients, as in our case.

Osteoarticular involvement is rare. A review of the American and English-language literature from 1924 to 1970 showed only 24 cases of sporotrichosis of the bone, with the tibia, fibula and tubular bones of the hand and foot being the most affected sites [6, 7]. However, unlike the case presented, the majority of osteoarticular occurrences described tend to cause monoarthritis, most often of the knee joint.

The radiologic findings of lytic lesions with involvement of metaphyses, associated with more aggressive lesions (ruptured cortical bone and expansion) in the hands and feet, indicate a main differential diagnosis of tuberculosis, paracoccidioidomycosis, chronic recurrent multifocal osteomyelitis, leukemia, primary multifocal osseous lymphoma or metastatic neuroblastoma.

Neuroblastoma is the most common source of metastatic bone disease. It occurs most often before age 5 years, typically with osteolytic lesions; preferential involvement of the metaphysis of long bones, vertebral collapse and involvement of the skull are common. Like neuroblastoma, leukemia is one of the main sources of metastases in childhood, usually affecting the metaphyseal region, with its principal feature being a permeative or moth-eaten presentation. Primary multifocal osseous lymphoma is rare in children, accounting for 2.8% to 4.2% of all non-Hodgkin lymphomas [8], mainly affecting adolescents; there is not involvement of lymph nodes and viscera for a minimum of 6 months from presentation and it may or may not affect the bone marrow. On radiographs, primary multifocal osseous lymphoma appears, in most cases, as osteolytic lesions, which may involve the epiphysis, and it has poor prognostic factors for rupture of the cortical bone, pathological fracture and soft-tissue mass.

Additionally, chronic recurrent multifocal osteomyelitis can progress to skin lesions, particularly palmoplantar pustulosis. The imaging findings on radiographs may show lytic lesions with sclerosis mainly confined to the metaphyses [8].

Among the possible infectious causes of the differential diagnosis, tuberculosis and fungi should be considered. Tuberculosis may present with skin lesions similar to the two patterns seen in this case, but with involvement preference of the vertebrae and with involvement of tubular and flat bones in only 10–15% of cases. Among the fungal diseases there is paracoccidioidomycosis (also called South American

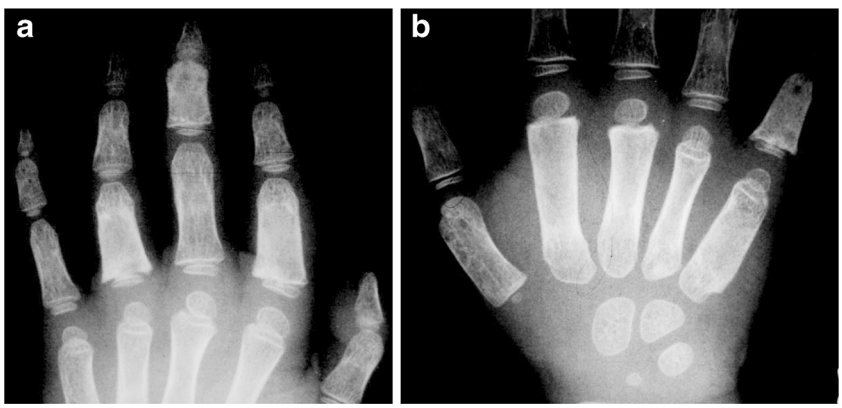

Fig. 5 Radiographs of the hands after 45 days of therapy with amphotericin B and itraconazole. Anteroposterior radiographs of the right hand (**a**) and left hand (**b**) show significant radiologic improvement, with sclerotic areas evident at the metadiaphyseal region of phalanges and metacarpal bones

blastomycosis), which can present with skin lesions and radiologic features identical to those of the reported case; paracoccidioidomycosis, however, is more common in the clavicle, ribs and bones of the forearm. Another possibility is the fungal disease actinomycosis; however this is a very rare cause of osteomyelitis.

A diagnosis that could also be hypothesized is Langerhans cell histiocytosis, a disease of the reticuloendothelial system that has its peak incidence at 2–6 years of age, with the skull being the most common site of bone involvement.

A rare form of osteoarticular sporotrichosis should be considered even when there are unusual manifestations, such as the combination of two patterns and the presence of well-defined metaphyseal lytic lesions with a halo of sclerosis, as presented here. This case was supported by the presence of skin lesions consistent with exposure to cats. An immunocompromised patient is not required to entertain this hypothesis. Bone involvement with *Sporothix schenckii* can occur in an immunocompetent host.

Conflicts of interest None

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