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CASE REPORT

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Subcutaneous Mucormycosis in an Immunocompetent Nigerian Child

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Abstract: A 13-year-old girl presented with a three–month history of generalized blisters on her body following a traumatic injury to her leg with a piece of wood while farming. The lesions were hemorrhagic, itchy and tender on pressure. The patient was non-diabetic, non-leukemic, non-neutropenic, and not previously on immunosuppressive drugs, nor was there any evidence of immunosuppression. Culture and microscopic examination of scrapings from the skin lesions revealed growth of a Mucor spp. She was initially placed on potassium iodide and responded well for about a month, then relapsed. The patient was successfully treated with itraconazole monotherapy.

Key Words: Mucormycosis, immunocompetent child, itraconazole monotherapy

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Introduction

Mucormycosis is a rare fungal infection seen most often in association with prolonged neutropenia. Important information has also been collected by clinicians and researchers regarding the association of zygomycosis with cancer (1), antibiotic or prednisone use (2), diabetes (1,2), deferoxamine and desferrioxamine therapy (3), transplantation (4), and combination of immunosuppressive therapies. Cases have also been observed in immunocompetent hosts and have been noninvasive or locally invasive without dissemination (5). With the development of diagnostic tools that allow earlier diagnosis, better surgical and antifungal interventions, and more sophisticated laboratory methods for identifying the offender agents, more patients are surviving these previously fatal infections. In the treatment of Mucor infections, amphotericin B is the drug of choice. *In vitro* testing has shown that both *Mucor circinelloides* and *Mucor hiemalis* are susceptible to amphotericin B (6). Other reported treatments include potassium iodide (1 g three times per day) for the treatment of subcutaneous lesions (7).

Case Report

A 13-year-old girl was referred to us for skin lesions (blisters) covering most of the body. The patient's illness had started three months prior to presentation with traumatic injury to the right leg while working on the farm. Initially, a blister had formed at the site of the injury, which later ruptured to discharge a yellowish foul-smelling fluid. The lesions had spread to the left leg and other parts of the body over the next two to three months. They were itchy and bleeding, especially those around the joints. No history of alcohol abuse, diabetes mellitus or leukemia was elicited. The patient was not receiving any immunosuppressive therapy. On physical examination, she was found to be mildly pale, afebrile and without peripheral lymphadenopathy. There were widespread hyperpigmented skin lesions with scaling, scab formation and foul-smelling serous discharge involving the lower and upper limbs, face, back, scalp and ears. The lesions were interspaced with hypopigmented patches and spots (Figures 1 and 2). They were hemorrhagic and varied in size. The rest of the physical examination was unremarkable.

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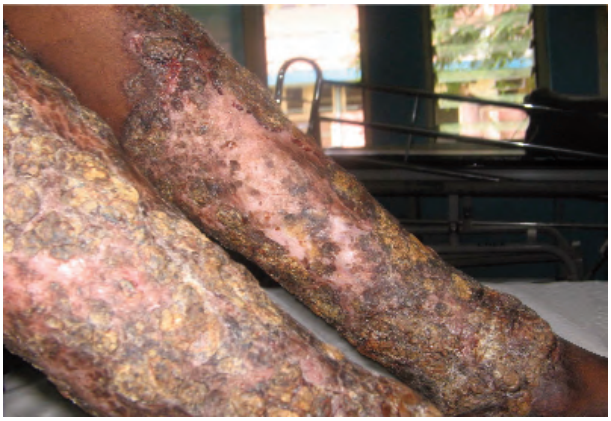

Figure 1. Skin lesions on left leg and left lower arm captured with digital camera.

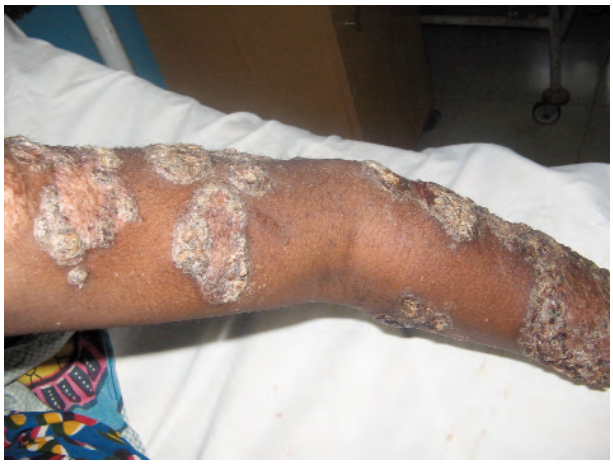

Figure 2. Plaque of skin lesions on left upper and lower arms captured with digital camera.

Initial tentative diagnoses were:

(i) Subcutaneous mycosis? Sporotrichosis. (ii) Skin tuberculosis? *Tuberculosis cutis verrucosa*.

Complete blood count revealed a hemoglobin concentration of 7gm% for which transfusion of packed red blood cells was performed. Chemical pathological, radiological and other hematological investigations revealed no other abnormality. Mantoux test was negative.

Scrapings from the skin lesions were dissolved with 10% potassium hydroxide on a clean glass-slide. Aseptate, ribbon-like hyphae of irregular sizes (10- 40 µm) and numerous sclerotic bodies were seen scattered throughout.

Cultures showed growth of a mold rapidly filling the Petri dish of Sabouraud's agar within 48 hours of incubation with white luxurious colonies. The optimum temperature for growth was 32–37°C; growth was poor at room temperature (25°C). The colonies were examined under light microscope.

The sporangia were globose with ellipsoidal columellae that were partially obscured by sporangiospores. Some sporangia were deliquescent releasing clumps of hyaline sporangiospores. The hyphae were broad and non-septate. Sporangiophores exhibited minimal sympodial branching. Rhizoids and stolons were absent (Figure 3).

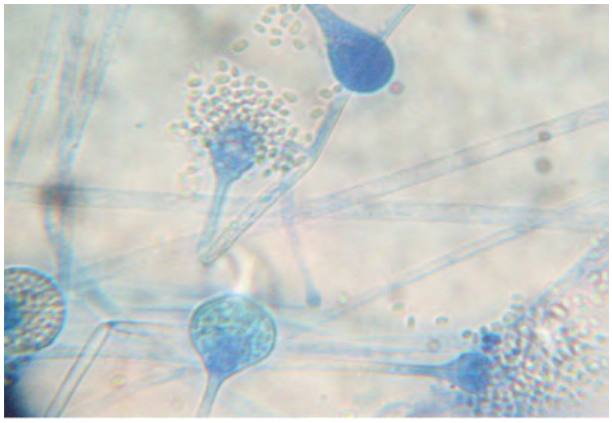

Figure 3. Lactophenol cotton blue (LPCB) mount of growth of *Mucur* spp, on Sabouraud's dextrose agar without cycloheximide. Sporangiophores are seen with ellipsoidal columellae and deliquescent sporangia releasing clumps of hyaline sporangiospores (400x).

The diagnosis of mucormycosis in this case was based on the presenting clinical features, cultural and microscopic characteristics and morphology of the fungus. The subspecies of the mucor has not been identified since immunological and molecular diagnostic investigations are yet to be done.

The patient was initially placed on potassium iodide and responded well for about a month, then relapsed. She was then successfully treated with itraconazole monotherapy at a dose of 200 mg twice a day for 7 months. She is currently on follow-up.

Discussion

Zygomycosis was first reported as a cause of human disease in 1885 (8). Unlike other filamentous pathogens that target immunocompromised hosts, zygomycosis organisms infect a broader and more heterogeneous population. Persons with no underlying conditions and patients with diabetes mellitus represented >50% of all infected patients (9). In a review of 929 cases of zygomycosis by Roden et al. (10), cutaneous zygomycosis was found to be the third most common type (19%) after sinus (39%) and pulmonary (24%) zygomycosis. Traumatic inoculation seems to be the means of introduction of the fungal spores into the skin in our patient. In this case, there was no underlying factor that could be incriminated as a risk factor in development of the disease. Although she was from the rural part of the country and was of low socioeconomic background, she was not malnourished. Another unusual experience with our patient was the favorable manner in which she responded to itraconazole monotherapy, as against the norm in which extensive debridement and amphotericin B therapy constitute the gold standard. This may be due to the fact that there was no evidence of dissemination of the infection to deeper tissues and organs of the body.

Measures to decrease the incidence of zygomycosis in patients at risk are difficult at best. There is no routine antifungal prophylaxis available, and with the low prevalence of zygomycosis, there is no real indication to provide it. The most common preventive interventions attempted involve modifications and controls in the environment that reduce the risk of exposure to airborne spores. Most of these control measures have focused on easily identified patients at risk, i.e., those expected to be profoundly neutropenic for prolonged periods. Transplantation and chemotherapeutic wards are often isolated with Hepa-filter treatment of the air supply and positive pressure to exclude the recruitment of dust into the ward (9). Adequate clothing and foot-wear are an important protective measure for farmers.

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