Case Report

Subcutaneous Zygomycosis Due to Basidiobolus ranarum: A Case Report from Maharastra, India

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Received 15 September 2010; Revised 3 November 2010; Accepted 22 November 2010

Academic Editor: Luis Eduardo Cuevas

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1. Introduction

Entomophthoromycosis (subcutaneous zygomycosis) is a sporadic subcutaneous infection that is largely restricted to tropical areas of Africa, Asia, and South America. It presents in two clinically distinct forms. Subcutaneous zygomycosis, caused by Basidiobolus ranarum and rhinofacial zygomycosis caused by Conidiobolus coronatus. Neither of these two forms occur preferentially in patients with underlying disease or defective immunity [1]. Entomophthoromycosis is characterized by the formation of firm and nontender swellings, generally on the extremities, trunk, and rarely other parts of the body [2]. Subcutaneous zygomycosis can also mimic soft tissue tumors [3]. We present a case of a 3-year-old Asian boy with a large, rapid-growing zygomycotic lesion of the left thigh which was initially misdiagnosed as a soft tissue tumor, emphasizing the importance of rapid and thorough diagnosis with a high index of suspicion, especially in endemic areas.

2. Case Presentation

A three-year-old male child was admitted with the complains of a gradually increasing painless swelling over the left thigh of 6 months duration. There was a history of trauma to the site three months prior to the onset of the present swelling. Local examination showed a 14 × 8 cm nontender indurated subcutaneous swelling on the left thigh over the anterolateral, medial, and posterior aspect. The swelling was firm in consistency with smooth and rounded edges and was freely mobile over the underlying structures. The overlying skin was lobulated and hyperpigmented and showed a 2 cm linear scar. There was no regional lymphadenopathy. Based on the history and clinical presentation, a diagnosis of soft tissue tumor was considered.

The Rest of the general and systemic examination was normal. Routine laboratory investigations revealed an elevated total leukocyte count (27,400/cmm). His HIV status was negative. X-ray of the thigh revealed a soft tissue swelling
without any bone involvement. Fine needle aspiration cytology of the swelling was suggestive of a spindle cell lesion with few inflammatory cells. The swelling was completely excised and sent for histopathological examination.

The excised specimen had a grey-white nodular cut surface. The haematoxylin- and eosin-stained sections revealed normal skin with subcutaneous tissue showing inflammatory granulomatous reaction with diffuse and dense infiltration by eosinophils, polymorphs, histiocytes, plasma cells, lymphocytes, and few multinucleated giant cells. The central portion showed fragments of aseptate hyphae surrounded by a peculiar eosinophilic mass and the Splendore-Hoeppli phenomenon (Figures 1 and 2). Periodic acid Schiff (Figure 3) and Gomori methenamine silver (Figure 4) stain for fungus showed few, broad, thin-walled, and aseptate fungal hyphae.

Culture on Sabouraud’s dextrose agar after three days of incubation showed furrowed creamy-brown radially folded colonies. Lactophenol cotton blue wet mount demonstrated aseptate hyphae and smooth-walled zygospores with characteristic conjugation beaks, which confirmed the fungus to be Basidiobolus ranarum.

After complete excision and in view of the histopathological report, the patient was started on oral potassium iodide 40 mg/kg per day. After one week of treatment with potassium iodide, oral itraconazole was added in a dose of 100 mg/day and the entire treatment continued for six months. The recovery was uneventful and the patient was discharged on the 10th postoperative day. After six months of followup there was no recurrence. The hematological examination showed total and differential counts within normal limits indicating complete recovery.

3. Discussion

In 1956 in Indonesia, Lei-Kian Joe described the first case of entomophthoromycosis in humans; the infectious agent was Basidiobolus ranarum. In 1965 Bras reported a case in a Jamaican native caused by Conidiobolus coronatus. Worldwide, there have been about 250 reported cases caused by Basidiobolus haptosporus. All of them affecting trunk and extremities, and usually seen in children. Approximately 160 cases caused by Conidiobolus coronatus have been reported
Most patients with entomophthoromycosis respond very well to oral potassium iodide therapy as well to azoles, particularly itraconazole [9].

To summarise, subcutaneous zygomycosis presents as a painless subcutaneous swelling usually in young age. It is usually seen in the natives of Asia, Africa, and South America. Demonstration of the asceptate fungal hyphae on histopathology and confirmation by culture clinches the diagnosis. Treatment with potassium iodide and azoles is the gold standard.

Our experience with this case highlights the importance of awareness and early recognition of this condition to prevent disfigurement produced by advanced disease, misdiagnosis, and unnecessary surgical intervention.

References
