

Subcutaneous Basidiobolomycosis Treated Successfully with Oral Itraconazole: A Case Report

Abstract

Basidiobolomycosis is an opportunistic fungal infection with a high morbidity and mortality rate. It is known to cause invasive disease in immunocompromised persons but it may produce only cutaneous or subcutaneous infections in an immunocompetent patient. Treatment is difficult due to its fulminant course and lack of effective anti-fungal drugs. But here, we report a rare case of Basidiobolomycosis detected in an immunocompetent patient—without any debilitating illness, which showed a complete response to itraconazole 200 mg daily without any surgical intervention.

Keywords: *Basidiobolomycosis, immunocompetent, itraconazole*

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Introduction

Zygomycosis is a group of fungal infections caused by fungi belonging to the class zygomycetes. The class includes two orders of medical importance: Mucorales and Entomophthorales. Entomophthoromycosis is caused by fungi of the order Entomophthorales. These are of two types: Subcutaneous zygomycosis (*Basidiobolus ranarum*) and Rhinofacial Zygomycosis (*Conidiobolus coronatus*).^[1,2] Basidiobolomycosis clinically manifests as chronic infection of subcutaneous tissue in an immunocompetent host involving trunk and extremities.^[3] Here we present a case of a young, immunocompetent male, with basidiobolomycosis who showed a steady, complete response to itraconazole 200 mg/day with a good long-term prognosis.

Case Report

An 18-year-old male patient presented to the Dermatology outpatient department with chief complaint of swelling over forehead more on the right side, right peri-orbital area, right cheek, and chin, extending to involve both sides of neck and shoulder, for 1.5 years associated with generalized malaise for 6 months, weight loss of 8–10 kg in last 6 months and mild grade evening rise fever associated with chills, partially relieved by medication for 15 days. There was no history of

trauma, seizures, pain over lesions, or any other systemic complaints like jaundice, diabetes, and no sign and symptoms suggestive of immunosuppression. Cutaneous examination revealed diffuse, firm, non-tender, nodular asymmetrical non-fluctuant swelling present involving bilateral cheeks (more over the right side), both lips, nape of the neck, bilateral infra-orbital area, lower eyelids (more on the right side), and forehead [Figure 1a-c].

There was a single 2 × 1 cm² sized non-tender firm mobile lymph node in the right axillary region, and two 1 × 1 cm² sized non-tender firm mobile lymph nodes over the right inguinal region. A complete hemogram was showed peripheral eosinophilia. Rest all the baseline investigations including liver and renal function tests were within normal limits and viral markers were negative. His coagulation profile, Mantoux test, Electrocardiogram, and serum Immunoglobulin did not reveal any abnormality. Local part ultrasonography showed coarse, ill-defined soft tissue swelling with vascularity in the subcutaneous and intermuscular plane of face and neck (more over the right side). The CT Scan findings of the neck showed evidence of multiple, discrete, confluent, heterogeneously enhancing varying sized nodular lesions with adjacent non-enhancing hypodense areas noted

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Figure 1: Diffuse hard Non-tender, Nodular asymmetrical non-fluctuant swelling. (a) Bilateral cheek involving bilateral cheeks more on the right side. (b) Right cheek and over upper chest. (c) Nape of the neck and upper back

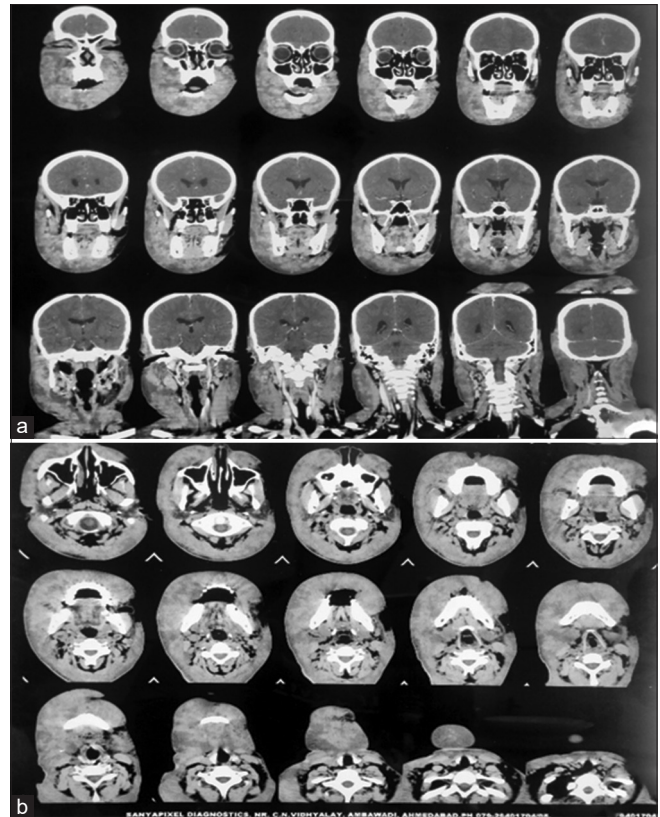


Figure 2: (a and b) CT scan of head and neck On presentation: multiple, discrete, confluent, heterogeneously nodular enhancing varying sized lesions with adjacent non-enhancing hypo dense areas within subcutaneous plane of face and neck, extracalvarial soft tissue on bilateral temporal region (more on right side)

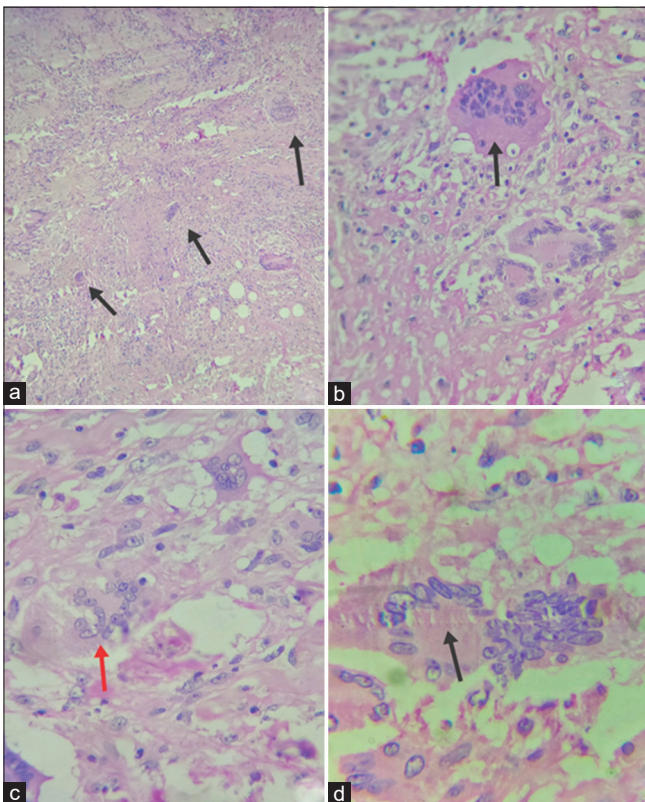


Figure 3: (a) Foreign body giant cell in the dermis (Black arrow) (H and E 100x), (b and d) Foreign body granuloma with non-septate hyphae and surrounding eosinophilic material (Black arrow- Splendore Hoespli Phenomenon) (H and E, b- 400x, d- 1000x) (c) Septate fungal hyphae (red arrow) (H and E, 400x)

within the subcutaneous plane of face and neck, the frontal region in mid-line, and extracalvarial soft tissue on the bilateral temporal region (more over the right side). These lesions are causing loss of fat planes, involving buccinators muscles and orbicularis oculi on both sides and right-side masseter muscles. Multiple enlarged lymph nodes were noted in bilateral submandibular upper mid-lower jugular posterior triangle levels and left parotid gland region largest measuring about 14 mm*12 mm [Figure 2a and b].

A skin biopsy was taken from the left clavicular region, left cheek, and upper back and sent for histopathological examination. Histopathology revealed subcutaneous infiltration of lymphocytes, histiocytes, and granuloma composed of epithelioid histiocytes and multinucleated giant cells, neutrophils, and eosinophils [Figure 3a-d]. Special staining with Periodic Acid Schiff (PAS) and Gomori Methamine Silver Stain (GMS) stain revealed intracellular and extracellular broad aseptate fungal hyphae. There were no acid-fast bacilli detected and no changes suggestive of neoplastic etiology on microscopic examination.

The clinical and histopathological findings suggestive of non-necrotizing granulomatous inflammation, suggestive of subcutaneous fungal infection, most likely

basidiobolomycosis. Itraconazole 200 mg daily was started. The patient was followed up every month for clinical improvement. Liver enzymes and cardiac monitoring were done at every visit. The therapy was continued for 9 months and stopped due to complete clinical and radiological improvement [Figures 4 a, b and 5a, b].

There has been no recurrence or new lesion even after 6 months of stopping treatment [Figure 6a, b].

Discussion

Subcutaneous basidiobolomycosis is endemic to rural areas of the inter-tropical zone, particularly Indonesia, Burma, India, particularly South India, and sub-Saharan Africa.^[4] The infection is caused by the filamentous fungus *Basidiobolus ranarum*, belonging to the class Zygomycetes and order Entomophthorales.^[5] Most likely, subcutaneous basidiobolomycosis results from inoculation of fungal spores into the dermis or subcutis through minor trauma such as insect bite, intravenous catheter, or even intramuscular.^[6,7] Usually, the disease starts as a hard, non-tender nodule on extremities or trunk which might progress and expand to form a larger hard, painless, non-tender swelling, that might ulcerate or even disseminate. Even though usually reported in immunocompromised individuals.^[8]

Our patient presented with a chronic subcutaneous infection of the right cheek, right supraclavicular area, and back without dissemination to internal organs, suggestive of basidiobolomycosis. Classically, the overlying skin is intact, with usually a bluish or purple hue (in light-skinned patients) at the point of maximum growth, and is movable over the swellings. Pruritus or a burning sensation may develop in severe cases due to secondary infections. For accurate diagnosis tissue biopsy is the ideal investigation. Culture is the gold standard for diagnosis.



Figure 4: Follow up after 9 months showing marked improvement in swelling. (a) Front view. (b) Side view

It is necessary to rule out soft tissue tumor, Hodgkin's lymphoma, Burkitt's lymphoma, mycobacterial infection, sporotrichosis, and A-V Malformation which may present in a similar manner.^[9] Histopathologically, subcutaneous basidiobolomycosis is characterized by small foci of suppurative granuloma distributed all over the dermis and subcutis. Different types of cells including lymphocytes, histiocytes, plasma cells, multinucleated giant cells, and mainly eosinophils play a major role.^[10] The presence of eosinophilic infiltrate in the granuloma is so characteristic that it is also called an eosinophilic granuloma. Degranulation of eosinophils leads to the formation of an eosinophilic sheath (Splendore-Hoeppli phenomenon) surrounding aseptate or infrequently septate thin-walled hyphae. Hyphae are easily visualized with hematoxylin and eosin stain. The successful management of infections caused by basidiobolomycosis depends on early diagnosis, control, or reversal of any underlying disease and antifungal therapy.^[1,7] Most cases of basidiobolomycosis have been treated with extensive surgical debridement but in our case, the favorable outcome is largely attributable to itraconazole. Patients treated with itraconazole show complete resolution of the disease, giving a new, conservative and safer approach to the treatment of basidiobolomycosis.^[3]

To conclude, early diagnosis and treatment can prevent permanent disfigurement and sometimes surgical

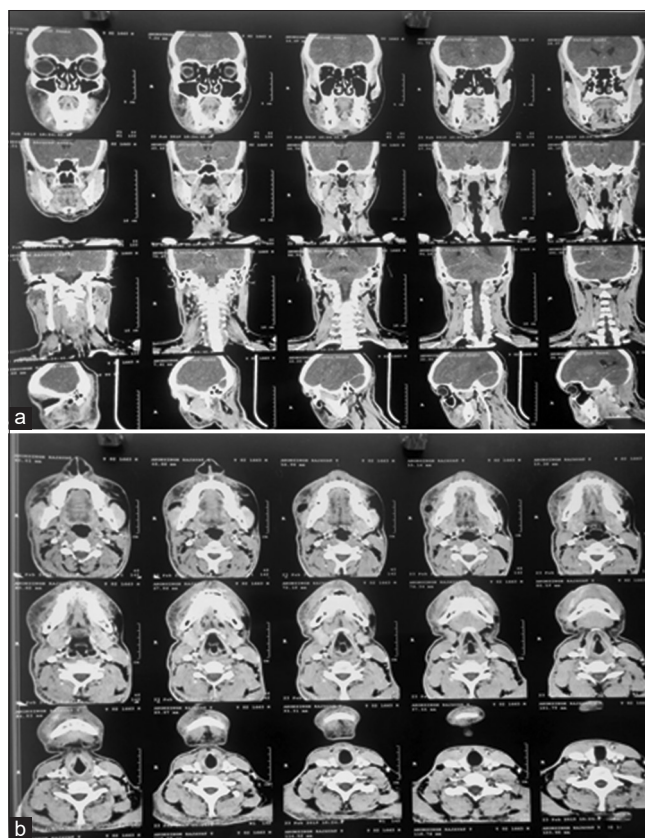


Figure 5: (a and b) Post-treatment CT scan of patient, after 9 months, showing marked reduction in heterogeneous nodular enhancing and non-enhancing areas within subcutaneous plane of face and neck



Figure 6: Follow up 6 months after stopping treatment (15 months from presentation) showing no residual swelling over face and neck. (a) Front view. (b) Side view

interventions in subcutaneous fungal infections. Subcutaneous zygomycosis must be considered as an important differential diagnosis in patients presenting with chronic facial swelling.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not

be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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