

# Basidiobolomycosis - Miss Me; It Hurts You

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## Abstract

Basidiobolomycosis is a subcutaneous infection of the limbs and trunk caused by *Basidiobolus ranarum*. It is one of the under diagnosed infection which usually occurs in tropical areas. High index of clinical suspicion is needed to diagnose this condition. We hereby report a case of basidiobolomycosis in a 25 year old male who presented to us with a non healing ulcer in left thigh. Biopsy showed granulomatous inflammation with fungal infection suggestive of phycomycosis. Fungal culture grew *Basidiobolus ranarum*. Patient was treated with oral itraconazole. The ulcer resolved completely after 3 months of treatment.

## Introduction

Zygomycosis is an infection caused by fungal agents belonging to the phylum Zygomycota. These are saprophytic fungi and are found ubiquitously in the environment. The class Zygomycetes includes two fungal orders: Mucorales and Entomophthorales. They have different pathogenic potentials; the order Mucorales primarily affects the immunocompromised patients causing invasive diseases like rhino cerebral or pulmonary mucormycosis with high mortality, where as the order entomophthorales, which include *Basidiobolus* and *Conidiobolus* genera, cause chronic infection in the subcutaneous tissue in the immune competent individual.<sup>1</sup> Basidiobolomycosis is caused by the fungus *Basidiobolus ranarum*. Here we present a case of subcutaneous basidiobolomycosis which was misdiagnosed as mucormycosis by histopathological examination.

## Case Details

A 25 year old male software engineer from Coimbatore was referred to us for intravenous amphotericin B, for the treatment of his non healing ulcer over the anterior aspect of left thigh as biopsy of the lesion was reported as mucormycosis. He had this lesion for the past 3 months which started as a painless nodular swelling (Figure 1A). He had consulted many clinicians earlier and was on multiple antibiotics, but his skin lesion progressively increased in size. He had no co-morbid illness and was apparently normal 3 month back. Ultra sonogram of the

thigh was suggestive of abscess in subcutaneous plane. He underwent incision and drainage (I and D) of the abscess which resulted in a deep cavitary lesion. Culture from the material during I and D was sterile. As the lesion was worsening (Figure 1B) he underwent another debridement. Gram's stain, AFB stain of the tissue didn't reveal any organisms and bacterial culture of the pus was sterile. Biopsy of the ulcer was performed and sent for histopathological examination (HPE). The HPE was reported as extensive ulceration and dense inflammation of dermis and subcutaneous area with fat necrosis and abscess formation composed of neutrophils and eosinophils along with thin walled broad aseptate fungal hyphae branching at right angles and surrounded by fibrosis and dense inflammation with giant cells. PAS stain was positive. Final report was foreign body granuloma with many fungal hyphae resembling mucormycosis. At this point we decided to re-evaluate the patient. On examination, he had a 3×1 cm linear non tender ulcer with surrounding induration on the left thigh (Figure 1C). His complete blood count showed total leukocyte count - 7400 (Neutrophils-50%, Lymphocytes-34%, Eosinophils-4%, Monocytes-7%), ESR - 24 mm in 1 hour. His blood glucose, renal function test and liver function test were within normal limits. Human Immunodeficiency virus 1 and 2 were

non-reactive. Since the history and clinical picture didn't fit with a diagnosis of mucormycosis (the morphology of the lesion, duration of the ulcer), a second opinion of the slide was sought. The histopathology description (Figure 2A, 2B, 2C) included the presence of granulomas in dermis and subcutis associated with the presence of numerous eosinophils. There were also scattered abscesses and eosinophilic material surrounding the fungal hyphae resembling Splendore-Hoeppli phenomenon. A probable diagnosis of subcutaneous phycomycosis was suggested with a comment that fungal culture was required for confirmation. Additional biopsy specimen was collected aseptically for fungal culture from the edge of the lesion. On Sabouraud's Dextrose Agar, furrowed creamy brown, heaped up, radially folded colonies grew after 3 days of incubation (Figure 3). Lactophenol cotton blue wet mount showed aseptate hyphae and smooth walled zygospores with characteristic conjugation beaks which confirmed the fungus to be *Basidiobolus ranarum* (Figure 4). After the diagnosis of basidiobolomycosis was confirmed, the patient was started on oral Itraconazole 200 mg once daily for 3 months. The ulcer healed slowly and complete resolution of the lesion (Figure 1D) was seen after 3 months.

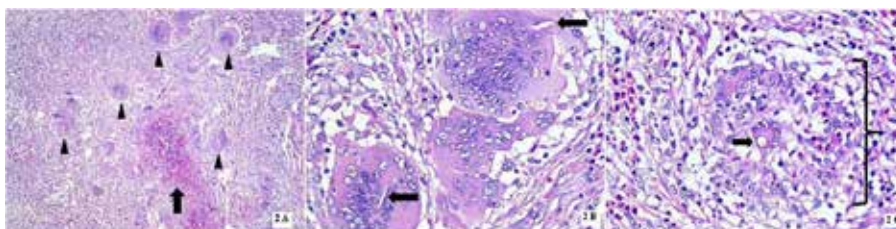
## Discussion

Basidiobolomycosis is caused by *Basidiobolus ranarum*, a saprophytic fungus present in soil, decaying fruit and vegetable matter as well as in the gut of amphibians and reptiles.<sup>2</sup> It can cause a variety of clinical manifestations including subcutaneous zygomycosis, gastrointestinal zygomycosis and occasionally an acute systemic illness. The subcutaneous form is a granulomatous infection of the skin and subcutaneous tissues characterized by the formation of firm, painless, disciform nodule generally on the

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**Fig. 1:** (A) Nodular swelling over thigh; (B) Painless ulcer after I and D; (C) Ulcer with induration after debridement; (D) Healed lesion after treatment



**Fig. 2:** (A) Low power view showing granulomas with many foreign-body type multinucleated giant cells (arrowheads) and Splendore-hoeppli phenomenon (arrow). H and E; (B) High power view showing giant cells with fragments of broad, aseptate fungal hyphae (arrows) in the cytoplasm. H and E; (C) High power view showing a granuloma (bracket) containing many Eosinophils and a giant cell with cross section of fungal hyphae (arrow) in the cytoplasm. H and E

thighs and buttocks in a “bathing suit” distribution.<sup>1</sup> It mainly affects young, male and immunocompetent persons. It is usually transmitted by minor trauma which may be through insect bite, intravenous catheter, or even intramuscular injection. Clinically it starts as small hardened swelling over the extremities and progressively increases in size to form a painless, non tender swelling. Macroscopically it closely mimics a soft tissue tumor, synovial sarcoma, Burkitt’s lymphoma or histopathologically mimics mucormycosis. The other differential diagnosis would be atypical mycobacterial infections, sporotrichosis and parasitic infections like oncocerciasis.<sup>3</sup>

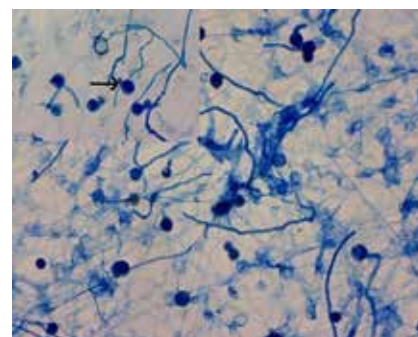
In our patient also it started as a small painless swelling and progressively increased in size. Even though the histopathology was reported as mucormycosis, the possibility of

mucormycosis could be ruled out by the absence of features such as angio/tissue invasion and necrosis; and slow growing nature of the lesion. Laboratory diagnosis is mainly based on histopathological features and culture of the organism which is the “gold standard”. Histologically, basidiobolomycosis is associated with eosinophilic infiltration; with short broad aseptate hyphae.<sup>4</sup> Some may be surrounded by an eosinophilic material which is called as ‘Splendore – Hoeppli’ phenomenon.<sup>5</sup>

Most patients with basidiobolomycosis respond well to oral itraconazole. The other treatment options are potassium iodide, trimethoprim-sulfamethoxazole. Some strains do not respond to amphotericin B. In the present case also the patient responded well to itraconazole and the ulcer got completely resolved after three months of treatment.



**Fig. 3:** Growth of basidiobolus ranarum on Sabouraud’s dextrose agar appearing yellowish grey rugose and smooth colonies with a waxy texture on the obverse end



**Fig. 4:** Lacto phenol cotton blue staining of basidiobolus ranarum. Non septate broad hyphae and zygospores with conjugation beaks seen (Insert - arrow) X 400 magnification

## Conclusion

This case is presented to insist on the importance of clinical correlation of histopathology reports along with proper sampling and appropriate microbiological test while treating rare infections.

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