



**Case Report:**

**Subcutaneous Phycomycosis in a Child**

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**Abstract:**

Subcutaneous phycomycosis is a rare entity. We hereby report a case of subcutaneous phycomycosis in 18 months old female child who presented with a painless, non-tender swelling on the thigh. Skin biopsy showed eosinophilic granuloma lying deep in the subcutaneous tissue, with sparse hyphae. Culture on Sabouraud's dextrose agar showed characteristic colonies. Patient was started on oral potassium iodide. The swelling was completely resolved after one month of treatment.

**Key Words:** Subcutaneous phycomycosis, Subcutaneous zygomycosis, *Basidiobolus ranarum*, Potassium iodide

**Introduction:**

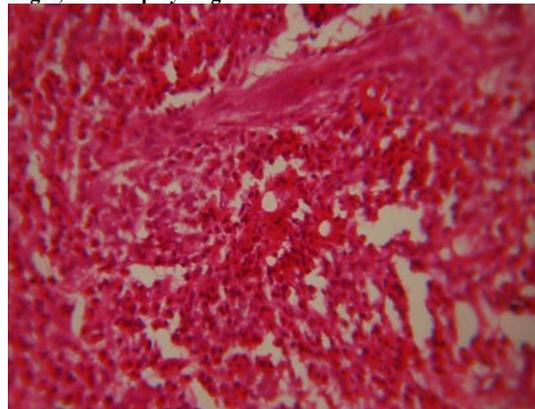
Subcutaneous phycomycosis (subcutaneous zygomycosis) is one of the deep fungal infections caused by zygomatous species which is normally found in the tropical countries. [1] *Basidiobolus ranarum*, a saprophytic fungus can cause a variety of clinical manifestations including subcutaneous zygomycosis, gastrointestinal zygomycosis and occasionally an acute systemic illness. Subcutaneous zygomycosis is the commonest presentation.[2] It is a granulomatous infection of the skin and subcutaneous tissues characterized by the formation of fluctuant firm and non-tender swellings, generally on the extremities and trunk.[3] It is predominantly a disease of early childhood.[4] Definitive diagnosis of subcutaneous phycomycosis can be made on histopathological study.[5] Oral potassium iodide can completely cure the disease.[1] In India, so far only a few cases have been described.

**Case Report:**

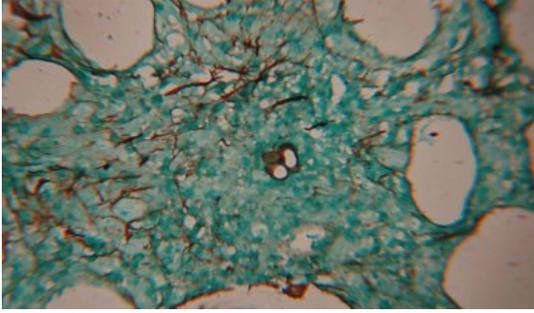
An 18 months old female was brought to dermatology clinic with complaints of a swelling over the left thigh for three months duration. Initially the swelling was small, measuring two centimeter in diameter. After two months, the swelling increased in size, measuring eight centimeter in diameter. Her general, physical and systemic examination and routine laboratory investigations were normal. X-ray of the thigh revealed a soft tissue swelling without any bone involvement. On examination, the swelling was non-tender with stony hard consistency, smooth rounded edges, raised up by finger underneath it. (Fig 1). The overlying skin was normal. A diagnosis of subcutaneous phycomycosis was suspected on the basis of clinical features.



**Figure 1:** The swelling over thigh, with smooth rounded edges, raised up by finger underneath it



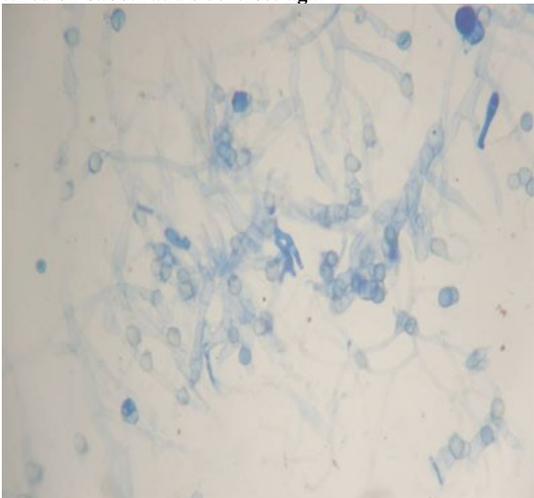
**Figure 2:** Photomicrograph shows granulomatous inflammation, with numerous eosinophils, lying deep in the dermis and subcutaneous tissue and largely replacing fat. (Haematoxylin and eosin X100)



**Figure 3: Photomicrograph shows occasional wide aseptate hyphae. (Gomori's methenamine silver stain X400)**



**Figure 4: Furrowed, creamy brown , heaped up, radially folded colonies after three days incubation at 25°C cultured on Sabouraud's dextrose agar**



**Figure 5: Lactophenol cotton blue wet mount of the culture growth showing numerous aseptate hyphae and numerous smooth walled zygospores with characteristic conjugation beaks**

Skin biopsy was taken. The Haematoxylin and eosin stained sections revealed granulomatous inflammation, with numerous eosinophils, lymphocytes and occasional giant cell lying deep in the dermis and subcutaneous tissue and largely replacing fat.(Fig 2) An eosinophilic material , suggestive of 'Splendore - Hoepplie' phenomenon was seen. Gomori's methenamine silver stain for fungus showed occasional wide aseptate hyphae. (Fig 3) Areas of fibrosis were noted. A 10% potassium hydroxide wet mount of the tissue revealed broad, thin walled and aseptate hyphae. Necrotic tissue from biopsy material was cultured on Sabouraud's dextrose agar, which showed furrowed, creamy brown, heaped up, radially folded colonies after three days incubation at 25°C.(Fig 4) On performing lactophenol cotton blue wet mount of the culture growth, numerous aseptate hyphae and numerous smooth walled zygospores with characteristic conjugation beaks were observed. (Fig 5) The fungus was identified as *Basidiobolus ranarum*. Patient was started on oral potassium iodide, three drops, three times a day and gradually increased upto five drops three times a day for one month. After one week of potassium iodide treatment, oral itraconazole in a dose of 100mg/day was added and was continued for one month. The swelling was completely resolved after one month of treatment.

#### **Discussion:**

Subcutaneous phycomycosis was first described in Indonesia in 1956.[4] It is the commonest clinical form of Basidiobolomycosis, and is endemic in South India.*Basidiobolus ranarum* is a saprophytic fungus present in soil, decaying fruit and vegetable matter as well as in the gut of amphibians and reptiles. It can cause a variety of clinical manifestations including subcutaneous zygomycosis, gastrointestinal zygomycosis and occasionally an acute systemic illness. Subcutaneous zygomycosis is the commonest presentation reported from many tropical countries including India.[2] However cases have not been reported from Maharashtra and north India. Traumatic implantation is probably the mode of entry like in other subcutaneous mycoses. In the past, clinical isolates of Basidiobolus were classified as *B. ranarum*, *B. meristosporus* and *B. haptosporus*. But recent taxonomic studies indicate that all human pathogens belong to *B. ranarum*.[2]

It is a granulomatous infection of the skin and subcutaneous tissues characterized by the formation of firm and non-tender swellings, generally on the extremities, trunk and rarely other parts of the body.[3] The disease usually occurs in children, less often in adolescents and rarely in adults. Males are much more frequently affected than females.[3] In the study by Lal S. et al, the sites of involvement included leg, thigh, buttock, shoulder, upper arm, thorax and abdominal wall and three patients had itching in the lesions and three patients were having fever.[6] In the present case there were no complaints, other than the swelling. The swelling may be lobulated with rounded edges and can be raised up by inserting the fingers underneath it.[1] In the present case also, the swelling could be lifted up by inserting the fingers beneath it. The possibility of mucromycosis could be ruled out by morphology of hyphae and absence of the features such as vessel involvement and thrombosis. Also the patient was in good general health with subcutaneous and painless swelling with intact overlying skin.

Laboratory diagnosis is based on histopathology. Histologically, Basidiobolomycosis is associated with eosinophilic infiltration, which was also the case in our patient. This has been postulated to be due to a predominant Th2 type of immune response with release of cytokines like IL-4 and IL-10 which in turn are helpful in recruiting eosinophils to the affected site.[2] The other histological features are extensive dermal and subcutaneous fibrosis and large zygomatous hyphae. The hyphae appear as short sections of broad hyphae without septae. Some may be surrounded by an eosinophilic material which is called

as 'Splendore – Hoepplie' phenomenon.[4] In the present case, the clinical and histopathologic features of skin lesion were characteristic of basidiobolus infection. Culture on Sabouraud's dextrose agar and lactophenol cotton blue wet mount confirmed the diagnosis of Basidiobolomycosis.

Most patients with Basidiobolomycosis respond very well to oral potassium iodide therapy as also to azoles particularly itraconazole.[2] In the present case also the patient responded well to potassium iodide and the swelling was completely resolved after one month of treatment. Prasad PV et al have observed that there was a relapse of the lesion in their patient after an excisional biopsy and have commented that surgery may hasten the spread of infection.[1]

To summarize, subcutaneous phycomycosis is a chronic fungal infection of the children, with a few reports from south India. The clinical and histopathologic features of skin lesion are characteristic of Basidiobolus infection. Clinical history, eosinophilic granuloma on histopathology and special stains for Basidiobolus help in clinching the diagnosis. Culture on Sabouraud's dextrose agar and lactophenol cotton blue wet mount are diagnostic. Surgical excision should be avoided as the patients respond well to oral potassium iodide.

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