Case Report

Basidiobolomycosis: Case Report and Literature Overview

Abstract

Basidiobolomycosis or subcutaneous zygomycosis or subcutaneous phycomycosis is a chronic granulomatous infection of skin and subcutaneous tissue, caused by a saprophytic filamentous fungus, *Basidiobolus ranarum*, clinically characterized by firm, painless subcutaneous swelling with smooth and rounded edges. Histopathological features include the peculiar Splendore–Hoeppli phenomenon. Culture on Sabouraud dextrose agar shows creamy white, heaped up, and furrowed colonies. This entity has been reported from tropical and subtropical regions of the world and the southern part of India. We report a case of Basidiobolomycosis in a seven-year-old girl from Eastern India, which was excised twice before presenting to us. We diagnosed the case as Basidiobolomycosis based on clinical features, histopathology, and culture findings, and treated it with itraconazole.

Keywords: *Basidiobolomycosis, Basidiobolus ranarum, itraconazole, subcutaneous phycomycosis, subcutaneous zygomycosis*

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Introduction

Zygomycosis designates a group of infections caused by fungi belonging to the class Zygomycota which includes two fungal orders, Mucorales and Entomophthorales. Mucormycosis is usually caused by Mucorales in immunocompromised patients, while Entomophthorales, a group of saprophytic fungi, affect immune-competent individuals causing entomophthoromycosis.[1]

Entomophthoromycosis is an uncommon sporadic, slowly progressive, and chronic subcutaneous infection that includes two distinct clinical forms, i.e., subcutaneous zygomycosis, caused by *Basidiobolus ranarum* and rhinofacial zygomycosis caused by *Conidiobolus coronatus*.[2]

Subcutaneous zygomycosis or subcutaneous phycomycosis or basidiobolomycosis is a chronic granulomatous infection of the skin and subcutaneous tissue in immunocompetent individuals, mainly restricted to tropical and subtropical countries with occasional reports from India.[1] We report a case of basidiobolomycosis diagnosed based on clinical features, histopathology, and culture findings. Our case highlights the importance of considering basidiobolomycosis early when immune-competent children present with unusual, painless, subcutaneous lesion, especially over the extremities.

Case Report

A seven-year-old girl child was brought to us with a painless swelling over the left knee joint since six months. Before presenting to us, she was treated by three physicians of different specialties. Initially, the lesion started as a small hard painless raised lesion over the dorsum of the left knee and gradually increased in size. The lesion was preceded by trauma at the site two months prior to onset. Parents consulted an orthopedic surgeon and the lesion was excised. After 1 month, a similar lesion again developed on the lower part of the anterolateral aspect of thigh just above the left knee. She was seen by a plastic surgeon and the lesion was excised and antibiotics were prescribed. The histopathology report of that time suggested nonspecific dermatitis. Two months later, similar painless swelling reappeared at the same site (dorsum of left knee) associated with fever and she was taken to a pediatrician. After one month of treatment, when no improvement was observed, the child was referred to us. On examination, an erythematous, ill-defined, nontender, and indurated swelling with smooth and rounded edges of size 5 cm × 4 cm with crusting

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over the surface was noted on the dorsum of the left knee joint [Figure 1]. The swelling was freely mobile and was not attached to the underlying structures. The lesion could be lifted up easily by inserting a finger beneath. The overlying skin was erythematous with one linear hypopigmented scar of length 4 cm on the left knee and one transverse scar of length 2 cm was found just above the left knee. There was no regional or peripheral lymphadenopathy. General and systemic examination revealed no abnormality. We considered a differential diagnosis of subcutaneous zygomycosis, lupus vulgaris, and soft tissue tumor. Routine hematological and biochemical investigations were normal, chest X-ray showed no abnormality, and Mantoux test was negative. Incisional skin biopsy specimen was sent for histopathological examination and fungal culture. Histopathology revealed inflammatory granulomatous reaction with dense and diffuse eosinophilic infiltrate and few multinucleated giant cells. The central portion showed fragments of broad, aseptate hyphae with peculiar eosinophilic material around the hyphae (Splendore–Hoeppli phenomenon) [Figure 2]. Periodic acid Schiff [Figure 3] and Gomori methenamine silver stain [Figure 4] showed thin-walled, broad, and aseptate fungal hyphae. Fungal culture on Sabouraud dextrose agar at 30°C after three days of incubation showed creamy white, heaped up, and furrowed colonies which identified the fungus as *Basidiobolus ranarum* [Figure 5]. The child was started on antifungal therapy with itraconazole at a dose of 4 mg/kg (100 mg/day) for 12 weeks. The lesion healed completely after two months of treatment [Figure 6]. However, she was advised to continue the treatment for 12 weeks. The treatment period was uneventful. She was followed up for six months and there was no recurrence.

Discussion

Entomophthoromycosis in humans caused by *Basidiobolus ranarum* was first described in Indonesia in 1956.[3]

B. ranarum is a saprophytic fungus present in soil, decaying fruit and vegetable matter, and the gut of amphibians, reptiles, bats, and fish. The mode of infection is exactly not known but it is assumed that

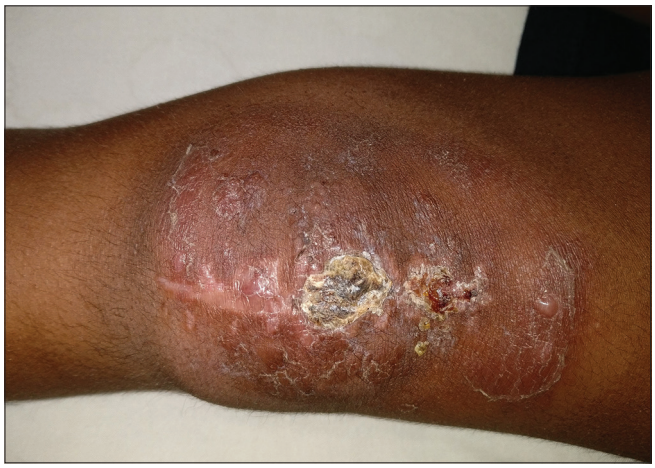

Figure 1: Erythematous, ill-defined, nontender, and indurated swelling with smooth and rounded edges with crusting and scaling over the surface

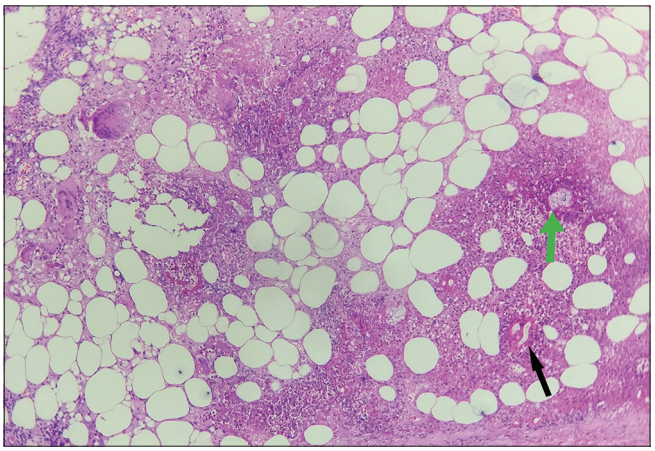

Figure 2: foci of granuloma (green arrow) and thin walled, broad aseptate fungal hyphae (black arrow). [H & E, 40×]

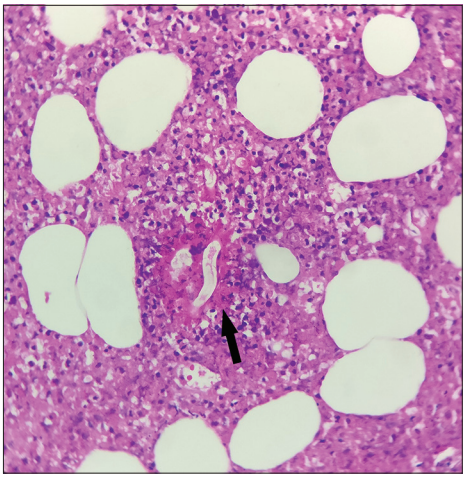

Figure 3: Broad, aseptate hyphae surrounded by bright eosinophilic material with a background of dense eosinophilic infiltrate "Splendore–Hoeppli" phenomenon. (PAS, 400×)

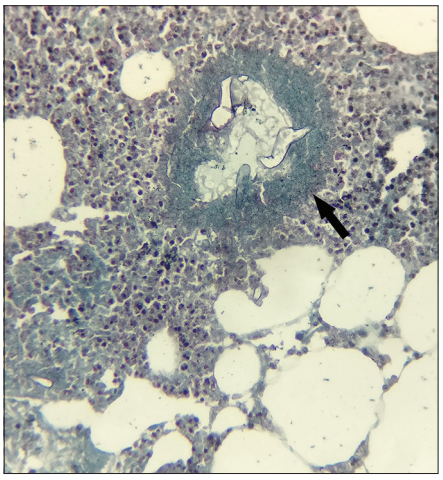

Figure 4: Aseptate fungal hyphal filament surrounded by inflammatory infiltration on Gomori methenamine silver (GMS) stain (GMS, 400×)

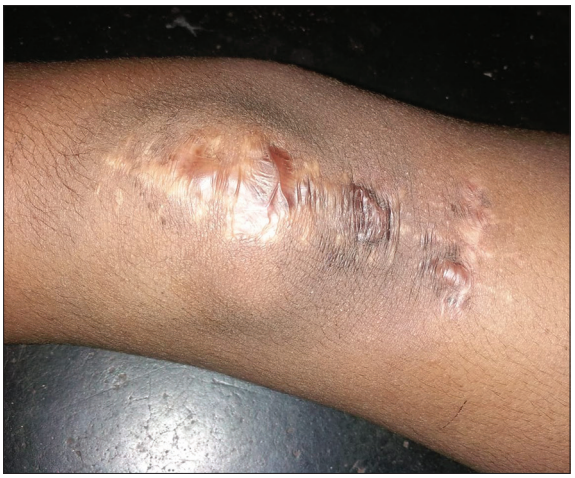

Figure 5: Creamy white, heaped up, and furrowed colonies on Sabouraud dextrose agar

Figure 6: Healed lesion after two months of treatment. Note two scar marks indicating previous surgeries

transmission may occur by implantation of spores of the organism via minor trauma such as insect bites, thorn prick, or by inhalation of spores which often goes unnoticed.[1] It can cause a variety of clinical manifestations including subcutaneous zygomycosis, gastrointestinal zygomycosis, and occasionally an acute systemic illness.[4]

Although the organism is found worldwide, the disease is prevalent in tropical and subtropical regions of the world. Few cases of Basidiobolomycosis have been reported from the southern part of India.[5-7] A summary of the published case reports of basidiobolomycosis from India has been elaborated in Table 1.

Basidiobolomycosis more commonly occurs in children, less often in adolescents, and rarely in adults. Males are more commonly affected than females.[1] Clinically, it manifests as a well-circumscribed firm, nontender, subcutaneous swelling on extremities (leg, thigh, buttock, shoulder, and upper arm)[8] or trunk, which if untreated may spread locally circumferentially or proximally and distally but systemic dissemination is extremely uncommon. The lesion is usually freely mobile and not attached to underlying fascia or muscle. The swelling may be lobulated, edges are smooth, rounded, and fingers can be easily insinuated under the swelling, virtually lifting it up. The overlying skin may be normal, erythematous, edematous, or hyperpigmented, but ulceration is rare.[1,8] Lymph nodes are usually not involved. The underlying bone is rarely involved. Absence of systemic signs is a characteristic clinical feature.[1,3,8] Though it is a benign entity, rarely it may cause severe morbidity extending to neck, trunk, and rectum.[1]

This clinical entity may resemble other deep fungal infections like mycetoma and sporotrichosis, lupus vulagaris, soft tissue tumor, Burkitt's lymphoma, synovial sarcoma, and fibrosing panniculitis.

Although the clinical appearance of the lesions of Basidiobolomycosis is very suggestive, histopathology and culture can only make a definitive diagnosis. The typical histopathological feature is the presence of thin-walled, broad, aseptate hyphae surrounded by brightly eosinophilic material with a background of dense eosinophilic infiltrate, known as "Splendore–Hoeppli" phenomenon. Culture on Sabouraud dextrose agar at 25°C–30°C after three days of incubation shows creamy brown, furrowed, heaped up, and radially folded colonies. Lactophenol cotton blue wet mount shows large, broad vegetative hyphae, and thick-walled zygospores with beak-like appendages characteristic of Basidiobolus.[3,5] The hyphae usually stain positive with Gomori methenamine silver, Masson's trichrome, and PAS stain.[9] In addition to culture, the causative agent, *Basidiobolus ranarum* may be diagnosed in an immunodiffusion test by detecting immune response against the agent.[10]

Treatment of Basidiobolomycosis is not always successful. Potassium iodide (KI) and itraconazole are commonly used for treatment. Other drugs like trimethoprim-sulfamethoxazole, amphotericin B, oral azoles like ketoconazole, 400 mg per day have been used.[5,11] KI at a dose of 30 mg/kg/day as a single daily dose or divided into three doses has been used. Itraconazole at a dose of 100–200 mg daily has been tried. Approximately, 6–12 months of treatment is usually needed with itraconazole.[12] In the present case also, the patient responded very well to itraconazole and the swelling completely resolved after two months of treatment. The role of surgical resection is controversial and surgery may hasten the spread of infection according to Prasad *et al*.[8]

Conclusion

This case report highlights the importance of high index of suspicion required to diagnose the condition in nonendemic areas and early diagnosis of this clinical entity to prevent

Table 1: A summary of published case reports of Basidiobolomycosis from India

Author	Age/sex	Site of lesion	Duration	Place from where the case detected	H/O trauma	Treatment given	Outcome
Roy AK *et al*. (2000)[11]	6 year/ Female	Bilateral buttocks	6 months	Kolkata	Present	Ketoconazole (Dose not explained)	Clearance of lesion within 4 months of treatment
Sethuraman G *et al*. (2001)[13]	3 year/ Female	Right upper extremity and chest	3 months	Tamilnadu	Not present	Oral saturated solution of potassium iodide	Complete regression after 3 months of therapy
Prasad PVS *et al*. (2002)[8]	18 months/ Male	Anterior chest wall	3 months	Tamilnadu	Not clarified by authors	Oral potassium iodide	Resolved completely after one month of treatment
Sujatha S *et al*. (2003)[5]	58 year/ Male	Left thigh	4 years	Pondicherry	Not present	Oral potassium iodide	Complete resolution within two months of therapy
Naniwadekar MR *et al*. (2009)[4]	18 months/ Female	Left thigh	3 months	Maharashtra	Not clarified by authors	Oral potassium iodide with oral itraconazole (100 mg/day)	Complete resolution after one month of treatment.
Thotan SP *et al*. (2009)[12]	10 year/ Male	Upper back	3 months	Manipal	Not clarified by authors	Lugol's iodine	Complete resolution after 3 months of treatment
Anand M *et al*. (2010)[14]	3 year/ Male	Left thigh	6 months	Maharashtra	Present	Combination of oral potassium iodide (40 mg/kg/day) and oral itraconazole (100 mg/day) for 6 months	Complete resolution
Verma RK *et al*. (2012)[15]	42 year/ Female	Neck and temporal region (right)	15 days	Chandigarh	Not clarified by authors	Intravenous amphotericin B deoxycholate (1 mg/kg bw) daily for one and half month and oral potassium iodide for two months	Complete resolution
Kumari PH *et al*. (2013)[6]	6 months/ Female	Left thigh	4 months	Andhra Pradesh	Present	Saturated solution of potassium iodide for 3 weeks	Not known
Jayanth ST *et al*. (2013)[16]	58-year/ Female	Right gluteal region	2 years	Chhattisgarh	Present (intramuscular injection)	Oral potassium iodide	Complete resolution in six months
Anaparthy UR *et al*. (2014)[17]	6 months/ Female	Left knee	4 months	Andhra Pradesh	Insect bite	Saturated solution of oral potassium iodide (KI)	Complete resolution after 8 weeks of treatment
Mondal AK *et al*. (2015)[18]	25 Year/ Female	Left arm	8 months	West Bengal	Not present	Saturated solution of potassium iodide	Responded remarkably within 2 months
Arora P *et al*. (2015)[19]	2½ year/ Male	Left buttock and upper thigh	6 months	New Delhi	Present (intramuscular injection)	Saturated solution of potassium iodide for 10 weeks	Complete resol ution
Chintagunta SR *et al*. (2016)[20]	12 year/ Male	Right thigh and buttock	6 months	Telangana	Not clarified by author	Itraconazole 100 mg twice daily for 12 weeks	Complete resolution
Rajan RJ *et al*. (2017)[21]	20 months/ Male	Left buttock	6 months	Jharkhand	Not present	Potassium iodide (10 mg/kg/day) and cotrimoxazole (10 mg/kg/day)	Completely resolved after 2 months of treatment
Nalini P *et al*. (2019)[22]	75 year/ Male	Left forearm and arm	6 months	Tamil Nadu	Not present	Saturated solution of KI	Complete resolution of the lesion at the end the 3rd month
Patro P *et al*. (2019)[23]	4 year/ Male	Right upper arm	4 months	Chhattisgarh	History of trauma could not be elucidated	Itraconazole syrup 9 ml daily (90 mg)	Lesion started to resolve within first month.

unnecessary surgical intervention, disfigurement produced by advanced disease, and unnecessary psychological trauma to the patient.

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Conflicts of interest

There are no conflicts of interest.

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