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Case Report

Subcutaneous Zygomycosis: A Case Report

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Abstract

Subcutaneous Zygomycosis is a rare opportunistic fungal infection caused by Basidiobolus ranarum. The disease occurs due to inoculation of fungal spores into dermis or subcutaneous tissue probably due to minor trauma. It presents usually in healthy individuals, especially in children or adolescent age group as a firm, painless, nodule or sinus on the trunk or the extremities and if untreated can spread locally and enlarge in size. Definitive diagnosis of subcutaneous zygomycosis is made by physical examination together with pathologic and microbiologic evaluation. Extensive surgical debridement along with pharmacological agents like potassium iodide, trimethoprim sulfamethoxazole, amphotericin B, oral azoles or potassium iodide combined with oral azoles have been used to treat subcutaneous zygomycosis. Though this entity is endemic in South India, only limited numbers of cases have been reported. Here we report a case of subcutaneous zygomycosis in a 2 year old boy presenting as recurrent left gluteal swelling.

Key words: Basidiobolomycosis, *Basidiobolus ranarum*, Zygomycosis

INTRODUCTION

Zygomycosis is an acute or chronic infection caused by several fungal agents belonging to the class Zygomycetes which includes two fungal orders: *Mucorales* and *Entomophthorales*, with extremely different pathogenic potentials. *Mucorales* affect only the immunocompromised causing mortality in excess of 60% in those affected, while *Entomophthorales* affects the immune competent and includes *Basidiobolus ranarum* causing subcutaneous zygomycosis and Conidiobolus coronatus causing rhinofacial zygomycosis.[1,2] Entomophthoromycosis is characterized by the formation of firm and non-tender swelling, generally on the extremities, trunk, and rarely over other parts of the body.[3] Although, the organism is found world-wide, only around 100 cases have been documented.[4] Here, we report a child with subcutaneous zygomycosis presenting with swelling in the left gluteal region recurring after excision biopsy.

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CASE REPORT

A 2-year-old boy from rural Tamil Nadu, presented with recurrent swelling over left gluteal region. It started as a small papule which gradually progressed in size over 3–4 months. The swelling was painless, non-itchy, not associated with discharge or changes over overlying skin except for mild hyperpigmentation. Routine blood investigations, blood sugar, and ultrasound examination were normal. He received repeated courses of topical and systemic antibiotics and anti-inflammatory agents but showed no improvement. Magnetic resonance imaging done showed ill-defined T1 hypointense/heterogeneously T2 hyperintense subcutaneous lesion 4.7 × 4.8 × 2.6 cm with diffusion restriction and areas of necrosis with no bony involvement. With a suspicion of soft-tissue tumor, excision biopsy of the entire swelling was done which was reported as panniculitis.

Child presented to us with prompt recurrence of the swelling within a month of complete excision at same site 1–2 cm above the previous lesion scar [Figure 1]. It was progressive, 4 × 5 cm in size at presentation, non-tender, indurated with smooth, and rounded edges over left gluteal area. Skin over the swelling was hyperpigmented and skin could not be pinched away from the swelling and had no evidence of punctum/discharge/sinus or regional lymphadenopathy. Scar of

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the previous excision was healed and unaffected. There were no constitutional symptoms, limitation of hip movement, recent weight loss, similar family history, animal bites or thorn pricks or trauma or vaccination in the site or any other significant medical history. Blood investigation revealed microcytic hypochromic anemia, normal erythrocyte sedimentation rate and serum immunoglobulin E. Review of the histopathological slide

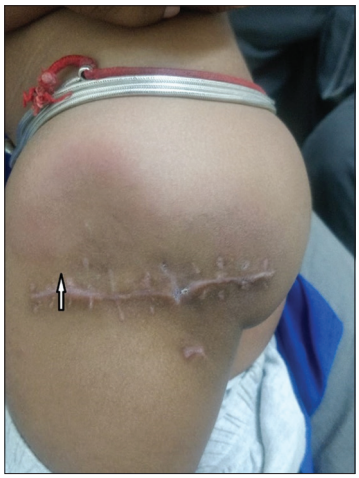

Figure 1

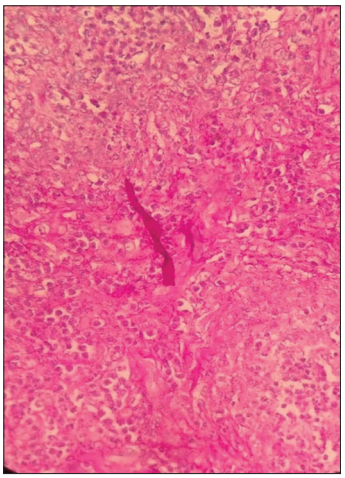

Figure 4

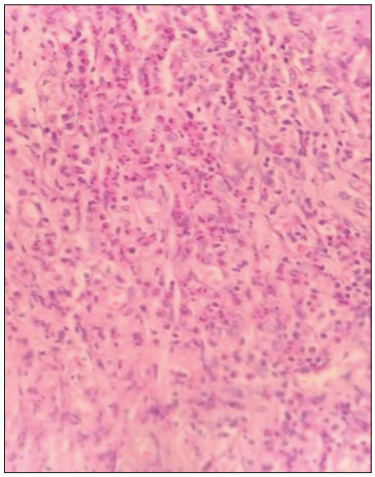

Figure 2

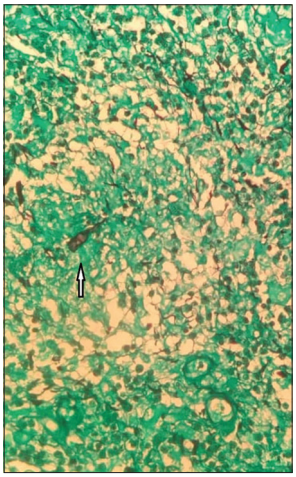

Figure 5

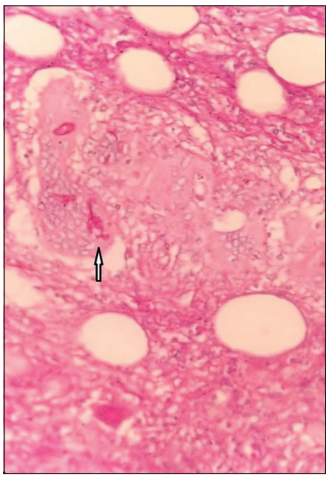

Figure 3

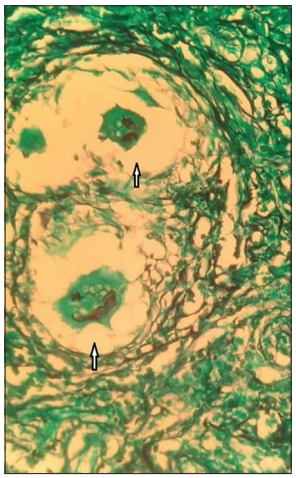

Figure 6

revealed plenty of eosinophils along with foreign body giant cells in hematoxylin and eosin staining [Figure 2]. There were foci of aggregates of epitheloid histiocytes and multinucleate giant cells forming granuloma which were occasionally suppurative. Few giant cells showed broad aseptate hyphae, highlighted by Periodic acid-Schiff stain [Figures 3 and 4] and Grocott-Gomori methanamine silver stain [Figures 5 and 6], and hence, a diagnosis of zygomycosis was made. Following the diagnosis, the child was started on potassium iodide. The child responded well to treatment given and showed good resolution by 6 weeks.

DISCUSSION

B. ranarum was initially described from animal and environmental sources as early as 1886. The first human case of subcutaneous zygomycosis was reported in 1956 in a patient from Indonesia.[4] Subcutaneous zygomycosis is a sporadic fungal infection that is largely restricted to tropical areas of Africa, Asia, and South America.[3] The disease is endemic in southern states of India. The disease usually occurs in children, less often in adolescent age group, and rarely in adults. Males are more frequently affected than females.[5]

The fungus *B. ranarum* is present in soil, decaying vegetable matter, and the intestines of amphibians, reptiles, fish, and insectivorous bats.[6] Subcutaneous zygomycosis results from inoculation of fungal spore into the dermis or subcutaneous tissue. Possible mode of transmission is minor trauma which may be through insect bite, intravenous catheter, or even intramuscular injection.[2]

Basidiobolomycosis occurs commonly in healthy individuals. *B. ranarum* causes a chronic infection of subcutaneous tissue, usually on the arms, trunk, and buttocks with most common presentation being on the thighs and buttocks in a "bathing suit" distribution.[4] It manifests clinically as a firm, painless, disciform nodule on the trunk or the extremities, which if untreated may enlarge in size and spread locally, but systemic dissemination is uncommon.[7]

The definitive diagnosis of basidiobolomycosis requires an excellent physical examination together with both pathologic and microbiologic evaluation.[6] Histologically, subcutaneous Zygomycosis is characterized by small foci of suppurative granuloma distributed all over the dermis and subcutis. Different types of cells including lymphocytes, histiocytes, plasma cell, and multinucleated giant cells contribute to the composition, but eosinophils play the major role, attributed to the release of IL4 and IL10 that help in recruiting eosinophils to the target site. The presence of eosinophilic infiltrate within the granuloma is so characteristic that it is also called as eosinophilic granuloma. Degranulation of these eosinophils leads to the formation of eosinophilic sheath (Splendore-Hoppelli phenomenon) surrounding aseptate or infrequently septate thin walled hyphae. Growth in standard fungal culture medium such as sabouraud dextrose agar is gold standard for confirming the disease if histopathology reveals doubtful results.[2] In addition to culture, diagnosis can be done by detecting an immune response in an immunodiffusion test developed for the diagnosis of basidiobolomycosis. This test has also been useful for monitoring the patients. Serology has been useful in making a diagnosis of disease even in the absence of culture.[8]

Extensive surgical debridement along with systemic antifungals is the standard treatment for cutaneous zygomycosis. Surgical debridement consists of complete resection of necrotic and infected tissue, often with a cuff of uninfected tissue. The wound must be closely monitored and at the first indication of disease progression, surgery must be repeated.[9] Pharmacological agents that have been used to successfully treat this infection include, most commonly potassium iodide, trimethoprim-sulfamethoxazole, amphotericin B, oral azoles, or potassium iodide combined with oral azoles. Treatment of Basidiobolus is not always successful, and no single drug has proved effective in the treatment of all cases.[6,10,11]

Basidiobolomycosis is a great mimicker of soft-tissue tumor, synovial sarcoma, and Burkitt's lymphoma. Hence, the possibility of misdiagnosing this disease as a neoplasm should be kept in mind.[2] It may also resemble tropical infections, fungal (Pythiosis and Sporotrichosis), parasitic (Filariasis and Onchocerciasis), and even bacterial infections (*Mycobacterium tuberculosis* and *Mycobacterium ulcerans*). Subcutaneous zygomycosis is a rare cause of soft-tissue infection and should be suspected in any atypical swelling, chronic non-healing sinuses and abscesses which are refractory to treatment.[12]

CONCLUSION

Subcutaneous zygomycosis is a rare opportunistic fungal infection caused my basidiobolus ranarum. Early diagnosis and awareness of this disease even in non endemic part of the country is very important to prevent misdiagnosis, disfigurement of the tissue, avoidance of unnecessary investigations and surgical interventions.

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