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Case Report

Subcutaneous basidiobolomycosis: An unusual fungus in immuno-competent children, vindicating longer duration of anti-fungal treatment

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ABSTRACT

Subcutaneous Basidiobolomycosis is a rare fungal infection caused by environmental saprophyte Basidiobolus ranarum. The disease is seen among immunocompetent individuals of age 18 months to 80 years, being more common in children. We report a case of Subcutaneous Basidiobolomycosis in a 10-year-old immunocompetent male child from Ahmednagar, Maharashtra who presented with complaints of right gluteal non tender, diffuse swelling, which was gradually progressive involving inner and back of thigh. The overlying skin was hyperpigmented with scaling at places. It was associated with complaints of difficulty in walking and voiding of urine. Patient was prescribed multiple medicines with no records available. All primary investigations were done along with a deep incisional biopsy for histopathology. MRI was suggestive of? lymphangioma? vascular malformation. Histopathology revealed panniculitis with dense collections of eosinophils. Few broad, aseptate fungal hyphae were seen camouflaged within the inflammatory infiltrate, surrounded by Splendore-Hoeppli phenomenon. The patient was treated with oral fluconazole for a period of one year with complete resolution of the symptoms. After one year of follow-up, the patient is clinically well. Clinically, this lesion presented as induration of skin and subcutaneous tissue thus mimicking a soft tissue sarcoma. However, histopathological examination clinched the diagnosis. Fungal infections are common in immuno-suppressed patients, and extremely uncommon in children. Since this is an exceptional rare fungal infection seen in immuno-competent children and young adults, it is often disregarded as a differential diagnosis.

1. Introduction

Basidiobolomycosis is a rare fungal infection caused by environmental saprophyte Basidiobolus ranarum, which belongs to order entomophthorales of class zygomycetes. It mainly involves skin and subcutaneous tissue and rarely the gastrointestinal tract.[1,2] Infection is mostly acquired by ingestion of infected food (eg. people with pica) or using contaminated papers for cleaning the skin (eg toilet paper). The infection is endemic in tropical and subtropical regions of Africa, Latin America, Middle East and Asia.[3] Infection is mostly common in immunocompetent paediatric population. The gold standard diagnosis for the infection are histopathology and fungal culture. The fungal culture on Sabouraud dextrose agar shows creamy white, heaped up, and furrowed colonies. Lactophenol cotton blue mount showed sparsely septate, broad hyphae, fragmented into short hyphal bodies and conidia of various sizes ,

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globose to pyriform shaped and smooth walled. The hyphae can also be appreciated on 40% KOH mount. The infection has excellent response to antifungal like Itraconazole and Fluconazole. We report a rare case of Subcutaneous Basidiobolomycosis in a 10 year old immunocompetent male child from Maharashtra (India) who presented with complaints of right gluteal non tender, diffuse swelling, clinically mimicking a soft tissue sarcoma.

2. Case History

A 10 year old male presented with ill defined, diffuse, non tender swelling with overlying hyperpigmented skin over right gluteal region since 6 months (Figure 1A). MRI right hip revealed well defined lesion in the subcutaneous plane of right gluteal region, posterior and medial aspect of right thigh suggestive of? lymphangioma? vascular malformation. The haemogram initially revealed mild increase in eosinophils (7.7%) followed by lymphopenia. There was also increase in value of ESR (31 mm at the end of an hour). The other blood parameters, blood sugar levels, renal and liver function tests were normal. The histopathological examination revealed skin covered fibrocollageneous and fibroadipose tissue with abscess formation composed of diffuse and dense inflammatory infiltrate of eosinophils, neutrophils, plasma cells, few lymphocytes and occasional multinucleated giant cells. Within the inflammatory exudate were scattered broad, stout, pauci-septate fungal hyphae, surrounded by bright eosinophilic material displaying Splendor – Hoeppli phenomenon**.** Foci of fat necrosis, proliferating capillaries and congested blood vessels were also seen. PAS (Periodic Acid Schiff) and GMS (Gomori's Silver Methanamine) special stains highlighted the fungal hyphae. All immunological tests for HIV, HBsAg, HCV and immunoglobulins were normal in our patient. He responded completely to oral fluconazole for one month.

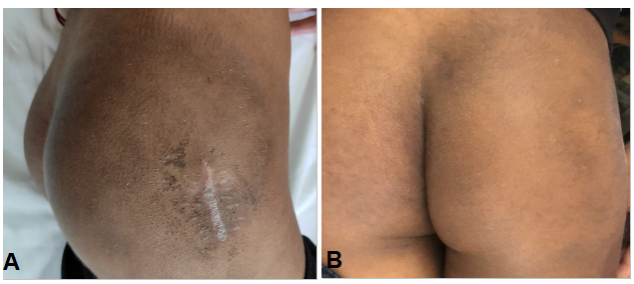

Fig. 1: Clinical picture of the site of the lesion; **A:** Pre-treatment: Induration, discolouration and thickening of skin over the gluteal region and upper thigh. Post-operative scar is noted; **B:** Post-treatment: follow up after one month of antifungal treatment

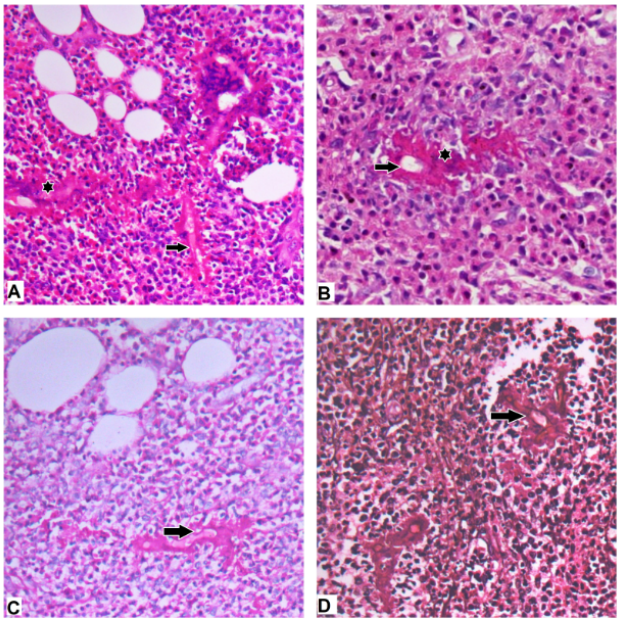

Fig. 2: Histopathology; A & B: H&E stain - Panniculitis with eosinophilia and Splendore Hoeppli phenomenon (star). Fungus is seen as broad, aseptate unstained wall (arrow); **C:** PAS stain D) Silver Methanamine stain

3. Discussion

Basidiobolomycosis is a saprophytic fungus which is more prevalent in tropical and subtropical regions of world including Asia.[4] It is common in male children, less likely in adults and adolescents and cause chronic infection of skin and subcutaneous tissue, rarely in gastrointestinal tract.[5] The affected skin or overlying skin of affected subcutaneous tissue is usually hyperpigmented, scaly or oedematous without any skin ulceration.[6,7]

The infection is usually seen in the subcutaneous fat tissues of limbs, gluteal region, chest or trunk of immunocompetent children.[8] Clinically, there is involvement of the skin and subcutaneous tissues, characterized by the formation of fluctuant, firm, and nontender swelling.There may be an increased erythrocyte sedimentation rate (ESR) and peripheral blood eosinophilia.[6] Contrast enhanced MRI of subcutaneous swelling shows marginated, irregular soft tissue mass localised in the skin and subcutaneous plane without involvement of underlying bone resembling any benign soft tissue lesion but with deeper infiltration at underlying superficial fascia.[9] Thus, this lesion may clinically mimic a sarcoma or tumour and lead to a misdiagnosis.

Histopathology and fungal culture are definitive diagnosis for the infection.[6] Histopathological examination reveals skin covered tissue infiltrated by acute and chronic inflammation with broad, stout, pauci-septate to aseptate fungal hyphae displaying Splendore – Hoeppli phenomenon

similar to our findings (Figure 2A&B)**.** Splendore Hoeppli phenomenon serves a clue to look for fungal elements, since it is brightly eosinophilic and easily visible under low power examination, while the refractile, unstained fungal cell wall may be difficult to identify at the first look. Focal areas of necrosis with granulomatous inflammation and zygospores may also be seen. Special stains like PAS (Periodic Acid Schiff) and Silver methanamine stains highlights fungal hyphae (Figure 2C&D)

Longer duration of therapy with anti-fungals is usually warranted, since these are resistant to treatment and can recur early. Our patient was treated with fluconazole for a duration of one year and responded well with no evidence of residual induration, swelling or pain. At the end of one year, he is well and free of disease (Figure 1B). It is important for the clinicians as well as the pathologists to be aware of this rare fungus since prompt treatment with antifungals is curative. We emphasize that this fungus affects predominantly young children and adolescents with immune-competent status and should be included in the differential diagnosis of soft tissue swellings with panniculitis.

4. Conflict of Interest

The authors declare that there are no conflicts of interest pertaining to the publication of this paper.

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None

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