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Subcutaneous Basidiobolomycosis in immunocompetent child

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Abstract

Subcutaneous basidiobolomycosis is rare deep mycotic infection but it has been reported to be endemic in certain rural and tropical areas.

A 2.5 years old girl child resident of rural area with low socioeconomic status presented with multiple lesions on buttock area on and off since 3 months. The lesions first appeared as a nodular painless swelling which gradually enlarged and then ulcerated. Local examination revealed multiple deep seated ulcers with black centre surrounded by healing margins and scaling of skin. Inguinal lymph nodes were enlarged. WBC counts and eosinophil count were raised. KOH mount revealed hyaline, thin walled, aseptate mycelia and yeast form which was suggestive of basidiobolus ranarum. Characteristic Splendore-Hoeppli phenomenon was observed on H & E stain. Patient was managed using topical application of potassium iodide for initial two weeks along with oral itraconazole (5 mg/kg daily) which was continued for 12 weeks.

Keywords: basidiobolomycosis, subcutaneous nodule, eosinophilia, ulcer

Introduction

Basidiobolus species are group of fungi belonging to class Zygomcetes, order Entomophthorales. These fungi typically causes gastrointestinal and subcutaneous lesions in trunk or limbs in immunocompetent and healthy individuals.[1] Though subcutaneous basidiobolomycosis is rare, it has been reported endemic in certain rural and tropical areas in Indonesia, India, particularly South India, Burma, and sub-Saharan Africa.[2] The fungi are present in decaying vegetable matter, soil, intestine of certain insects, reptiles and bats.[3] The precise mode of transmission of these infections is unknown but it has been speculated to be acquainted via dermal route following bite of an insect. Also it may occur by direct inoculation in the perineum due to contact with decaying vegetable matter.[4] Thus this theory explains buttocks, thigh and perineum to be most affected sites.

Children are most frequently affected as compared to adolescents and adults and higher proportion of cases have been documented in males as compared to females.[5]

Here we have described a case of subcutaneous basidiobolomycosis presenting as deep seated recurrent lesions in the buttock area of a young female child.

Case presentation

A 2.5 years old girl child presented to our OPD with multiple lesions on left buttock area on and off since 3 months. She was a resident of rural area near Bhopal, and belonged to low middle socioeconomic status. The lesions first appeared as a nodular painful swelling which gradually enlarged and then ulcerated. These lesions were not associated with any other complaints such as fever, weight loss, cough, loose stools etc. Her immunization was appropriate for age. On examination, weight and height were appropriate for age. Here development was normal. Locally, multiple deep seated ulcers, largest being approximately 2 x 3 cm^2 on left buttock was observed with black centre surrounded by healing margins and scaling of skin. Temperature of the affected area was normal. Minimal erythema and hyperpigmentation was observed surrounding the lesions. Multiple scars of healed lesions were also present. Her inguinal lymph nodes were enlarged to approximately 2 cm. The ulcers were not associated with any discharge or swelling of the adjacent area. Systemic examination revealed no abnormality.

Complete hemogram revealed reduced hemoglobin level (9gm/dl), Total WBC counts (17000/ cu mm) and eosinophil count (55% of total WBC) were raised. HIV and HBsAg were negative. Initially since eosinophilia was present, the child was treated on the line of filariasis with DEC. Meanwhile her KOH mount was conducted which revealed hyaline, thin walled, aseptate mycelia and yeast form which was suggestive of basidiobolus ranarum. Skin biopsy was conducted and sample was sent for histopathological examination which was suggestive of dense subepithelial inflammation predominantly composed of eosnophils and plasma cells. Characteristic Splendore-Hoeppli phenomenon consisting of sparsely septated hyphae surrounded by granulation tissue, eosinophils, lymphocytes and giant cells was observed on H & E stain.

Patient was managed using topical application of potassium iodide for initial two weeks along with oral itraconazole (5 mg/kg daily) which was continued for 12 weeks. Patient responded well to the treatment and lesions healed without recurrence.

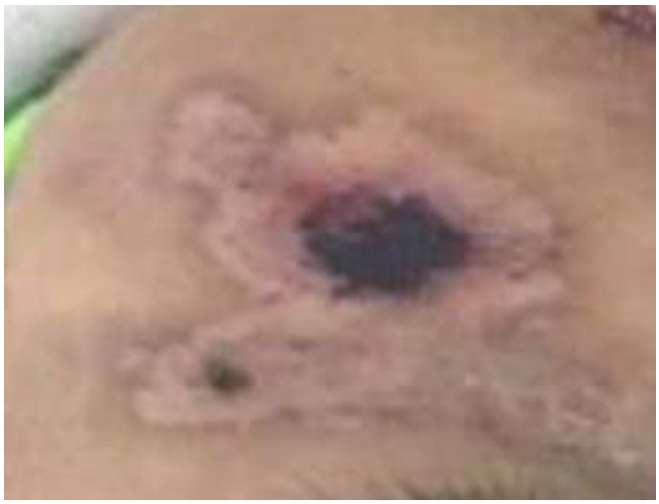

Figure 1: Lesion at the time of presentation

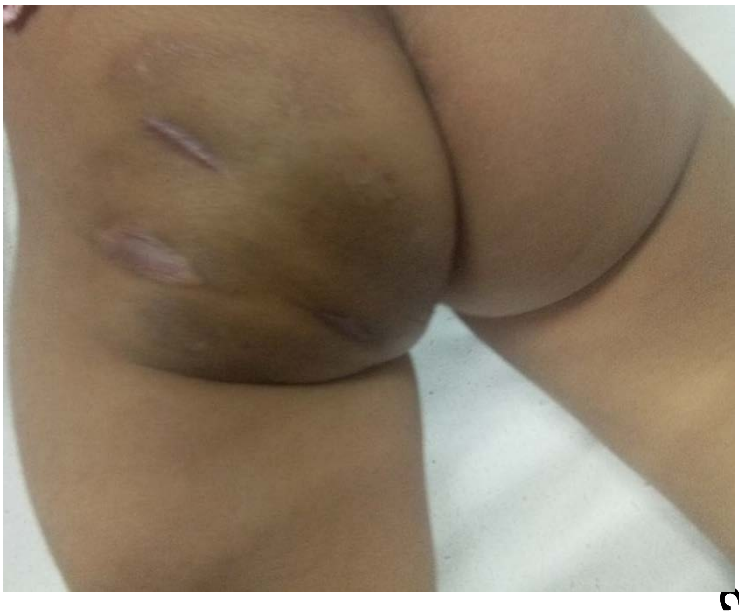

Figure 2: Healed lesion after 8 weeks of therapy

Discussion

Subcutaneous basidiolobomycosis is caused by Basidiobolus Ranarum which is zygomycosis fungi. The infection proceeds through following three stages depending upon the virulence of fungi and competence of the host- at initial stage, the infection remains confined to subcutaneous or cutaneous tissue, then the fungus may invade muscule and bones, at the final stage, it may cause disseminated infection by invading blood vessels.[6]

The mode of transmission of B ranarum remains unknown, however it is postulated that the infection is acquired through the skin after the bite of an infected insect or it may also occur following cleaning of anal or perianal area with the decaying leaves on which fungus are known to grow.[7]

Infection is more predominant in males as compared to females, however in present case scenario, we reported the case in 2.5 year old female. The commonly afflicted areas include perineum, buttocks, trunk and thighs. **Sackey A et al (2017)** in their case scenario documented that the typical presentation of Basidiolobomycosis is a firm subcutaneous nodule which is painless and indurated. The lesion then gradually enlarge with clear margin.[8]The lesion in present case appeared on buttock as a nodular painful swelling, the lesion enlarged gradually and ulceration was also noted. These lesions in our case were recurring as evident from healing scar marks surrounding the affected area. The risk factor associated with development of such ulcer could not be identified.

The clinical features of Basidiobolomycosis may mimic the features of certain tropical infections which presents with subcutaneous lesions. As the infection is rare, the diagnosis usually depend upon vigilant observation and investigations. The differential diagnosis of Basidiobolomycosis include parasitic infections (filarial elephantiasis and onchocerciasis), other fungal infections (pythiosis and sporotrichosis), bacterial infections (Mycobacterium ulcerans), and other diseases such as Burkitt's lymphoma and soft tissue tumour.[9]

Investigation such as CBC may reveal eosinophilia, raised C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) levels.[8] In our case scenario, total WBC counts (17000/ cu mm) and eosinophil count (55% of total WBC) were significantly raised.

Definitive diagnosis can be established by microscopy and skin biopsy. Characteristic lesions on biopsy include broad, thin walled hyphae together with acute and/or chronic inflammatory cell infiltrates and presence of Splendore-Hoeppli phenomenon which consist of sparsely septated hyphae surrounded by granulation tissue, eosinophils, lymphocytes and giant cells. Such characteristic lesions were observed in our case. Culture as well as immunodiffusion test can also be helpful in making the definitive diagnosis.[10]

Basidiobolomycosis can be effectively managed using antifungal drugs such as ketoconazole, itraconazole and fluconazole. Potassium iodide and co-trimoxazole are also helpful to control secondary bacterial infection at the site of ulcer. However no single drug has proved effective but a combination therapy may help in achieving cure. Undertreatment may lead to recurrence or resistance to the drugs, thus it is advised to continue treatment till 1 month after the lesions have healed completely.[8]

Conclusion

Basidiobolomycosis, a form of deep fungal infection, though uncommon may present as subcutaneous infection. The common site of involvement being perineum and gluteal region in immunocompetent host.

The child in our institute presented with recurrent painless nodular swelling in buttock area which was initially misdiagnosed and treated as a case of filariasis due to hypereosinophilia. As the infection is uncommon and awareness regarding the same is less, it should be considered in a child from area where hygienic conditions are suboptimal presenting with painless nodule, with/without ulcer in the usually afflicted sites like perineum, buttocks or thigh.

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