Original Research Paper

Volume - 12 | Issue - 01 | January - 2022 | PRINT ISSN No. 2249 - 555X | DOI : 10.36106/ijar

Dermatology

SUBCUTANEOUS ZYGOMYCOSIS PRESENTING WITH MULTIPLE SWELLINGS

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ABSTRACT Subcutaneous mycoses are a heterogenous group of fungal infections of the dermis and subcutis. They affect population in rural communities, often in humid, tropical, or subtropical regions of developing countries. They are directly inoculated into the dermis or subcutaneous tissue through a penetrating injury from vegetative materials.30 year old homemaker presented with two discrete painful hyperpigmented swellings with no surrounding satellite lesions, gradually increasing in size, on right thigh of six months duration. There was a history of penetrating trauma. No comorbidities were present .Histopathology showed septal and lobular panniculitis with foreign body granuloma. Fungal stains showed broad hyphae and Basidiobolus species growth was observed in Sabaroud dextrose agar. A diagnosis of subcutaneous zygomycoses was made and treatment with Itraconazole and potassium iodide was started, to which she responded well within 3 months of therapy. Histopathological examination is reliable and less time consuming. But culture is the gold standard investigation for confirming the diagnosis. The positivity in culture ranges from 40 to 60%, hence histopathology is the main tool for diagnosis. Treatment with itraconazole and potassium iodide has produced excellent results.

KEYWORDS : Basidiobolus species, Itraconazole, Potassium iodide

INTRODUCTION: Subcutaneous zygomycosis or subcutaneous phycomycosis or basidiobolomycosis, caused by Basidiobolus ranarum, is a chronic granulomatous infection affecting skin and subcutaneous tissue in immunocompetent individuals living in tropical and subtropical countries.

It is important to consider subcutaneous zygomycosis early in diagnosis when immunocompetent patients present with painless subcutaneous lesions, especially on the extremities and start immediate treatment with potassium iodide, which is the drug of choice. Here we report a case of subcutaneous zygomycosis diagnosed based on clinical features, histopathology and culture, which responded well to treatment with potassium iodide and itraconazole.

CASE REPORT:

A thirty year old female was referred to Dermatology OPD with two gradually increasing painful swellings over the upper and lower part of right thigh of six months duration. The lesions initially started as pea sized lesions which gradually increased in size to the present configuration. There was a history of penetrating trauma on right thigh preceding the onset of lesions. No history of weight loss, loss of appetite and fever. No history of other comorbidities ruling out immunocompromised status. Dermatological examination revealed hyperpigmented tender subcutaneous nodules of size 9 x 7 cm (Figure 1) and 6 x 4 cm (Figure 2) on the medial aspect of lower right thigh and anteromedial aspect of upper right thigh respectively. The lesions were firm to hard on palpation and freely mobile over the underlying structures. The margins were well defined and the lesions were indurated with positive finger insinuation test. Patient was afebrile and there was non-pitting edema of right lower limb. There was no regional lymphadenopathy. Systemic examination was within normal limit.

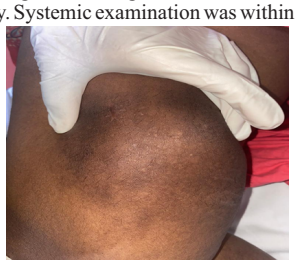

Figure 1 : Hyperpigmented tender subcutaneous nodule of size 9 x 7 cm present on lower thigh

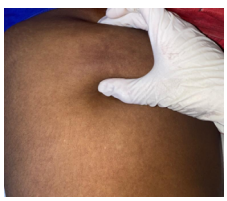

Figure 2 : Hyperpigmented tender subcutaneous nodule of size present on upper right thigh

Routine investigations were normal. MRI showed large irregular heterogenous poorly marginated lesions involving the dermis and subcutis with fascial infiltration over the anteromedial proximal thigh and medial aspect of the distal thigh with significant associated inflammatory stranding and subcutaneous edema. Biopsy samples were taken from both the lesions and sent for histopathology and culture. Histopathology showed septal and lobular panniculitis with foreign body granuloma and diffuse infiltrates of lymphocytes, plasma cells and eosinophils. Broad fungal hyphae was also noted. Culture showed luxurious, creamy white, membranous centrally heaped up radially folded colony of Basidiobolus species.(Figure 3)

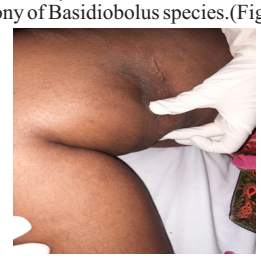

Figure 3 : Creamy white, heaped up furrowed colonies of Basidiobolus species in Sabaroud dextrose agar

After confirming the diagnosis as subcutaneous zygomycoses with histopathology and culture, the patient was started on saturated solution of potassium iodide and itraconazole with strict monitoring for signs of iodism and abnormalities in thyroid and liver function tests. The lesions responded remarkably within three months, with the lesion on lower thigh reducing to a size of 2 x 2 cm (Figure 4) and the upper thigh lesion almost disappearing completely leaving behind hyperpigmentation.

Figure 4 : Post treatment - Hyperpigmented subcutaneous nodule of size 2 x 2 cm present on lower part of thigh

DISCUSSION:

Zygomycosis is a group of fungal infections belonging to the class Zygomycota. It includes two fungal orders Mucorales and Entomophthorales. Mucorales causes mucormycosis in immuno compromised patients, whereas Entomophthorales causes entomophthoromycosis in immunocompetent patients.[1]

Entomophthoromycosis is an uncommon slowly progressive, sporadic and chronic infection. It has two clinical forms, subcutaneous zygomycosis caused by Basidiobolus ranarum and rhinofacial zygomycosis caused by Conidiobolus coronatus.[2]

Basidiobolomycosis was initially described by Joe et al in 1956. Emmons identified the causative organism isolated from these cases as Basidiobolus ranarum. It is the second most common deep mycosis in south India, the most common being mycetoma. It mainly affects children, particularly boys. It can cause subcutaneous zygomycosis, gastrointestinal zygomycosis and sometimes, an acute systemic illness extending to neck, trunk and rectum.[2,3]

The incubation period is not known. It has been suggested that the fungus enters skin through insect bites, accidental or self-inflicted wounds[4]. The infection mainly involves l imb girdles or proximal limbs. It presents as a painless, well circumscribed, freely mobile, firm to hard slowly growing subcutaneous mass with smooth, clearly defined borders. The mass can be raised up by inserting fingers underneath the lesion. It spreads by subcutaneous route. Satellite lesions develop at the advancing margins of the lesion. The overlying skin may be tense, edematous, hyperpigmented, ulcerated or desquamating.

Spontaneous resolution has been found to occur in a few cases. Differential diagnoses include mycetoma, sporotrichosis, lupus vulgaris and soft tissue tumor.

On histopathology, thin walled broad aseptate hyphae surrounded by bright eosinophilic material is seen with a background of dense eosinophilic infiltrate, known as

Splendore – Hoeppli phenomenon[1]. Culture on Sabouraud dextrose agar at 25 – 30 degree Celsius shows creamy brown, heaped up, furrowed and radially folded colonies after three days of incubation. Large, broad vegetative hyphae and thick walled zygospores with beak

like appendages, characteristic of Basidiobolus is seen in lactophenol cotton blue wet mount. Gomori methenamine silver, PAS and Masson's trichrome stains are used to detect the hyphae. Basidiobolus ranarum can also be confirmed by detecting immune response against the agent in an immunodiffusion test. The main characteristic features for diagnosis are slow spreading, painless, subcutaneous induration, eosinophilic granulomatous tissue on histology, presence of branched hyphae of phycomycoses, positive culture and negative on routine laboratory tests[5].

Potassium iodide and itraconazole are commonly used in treatment of subcutaneous zygomycosis. Potassium iodide has a direct antifungal action and it enhances myeloperoxidase activity and proteolytic activity[6]. It is supposed to localize at sites where the organisms are located[7]. KI at a dose of 30 mg/kg/day as a single daily dose or divided into three doses can be used. Itraconazole at a dose of 100 – 200 mg/day can be used. Approximately six to twelve months of treatment with itraconazole is usually needed[8]. Other drugs used include trimethoprim-sulfamethoxazole, amphotericin B, ketoconazole can be tried as second line agents. Surgery may hasten the spread of infection and its role is controversial.[9]

The occurrence of the disease in an adult female patient is a rare occurence as only three such cases have been reported from India. The occurrence of two discrete lesions is a rare feature in this condition.

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