ORIGINAL RESEARCH PAPER

Volume-8 | Issue-4 | April-2019 | PRINT ISSN No 2277 - 8179

INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

SUBCUTANEOUS PHAEOHYPHOMYCOSES CAUSED BY *EXOPHIALA SPINIFERA* IN AN IMMUNOCOMPETENT HOST- A RARE CASE REPORT

Medical Science

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ABSTRACT

Subcutaneous phaeohyphomycoses, a localised fungal infection of the deep dermis and subcutaneous tissues. It is caused by dematiaceous (darkly pigmented) fungi that are widely distributed in the environment. Clinically, these fungi cause diseases ranging from mild, superficial infections to fatal cerebral phaeohyphomycosis in otherwise healthy individuals.The most organisms commonly isolated in subcutaneous phaeohyphomycosis are *Exophiala* spp., *Bipolaris* spp. and *Phialophora* spp. We report a case of subcutaneous phaeohyphomycosis caused by *Exophiala spinifera* in an immunocompetent host.

KEYWORDS

Phaeohyphomycoses, Subcutaneous nodule,Dematiaceous fungi, Exophiala spp

INTRODUCTION

Phaeohyphomycosis is a term used to describe any infection with a pigmented mold, which also encompasses two ***specific*** syndromes—eumycetoma and chromoblastomycosis. A unique characteristic feature of these fungi is their ability to produce melanin within their cell walls and form yeast and/or hyphal-like structures in host tissues[1].

Most of these fungi cause localized subcutaneous infections after direct inoculation, but disseminated infection and serious visceral focal infections also occur, especially in immunocompromised patients. *Alternaria*, *Exophiala*, *Curvularia*, and *Wangiella* species are among the more common moulds reported to cause human infection[2]. We report a case of subcutaneous phaeohyphomycosis caused by *Exophiala spinifera* in an immunocompetent host.

CASE REPORT

A 50 year old farmer presented with a non tender, fluctuant, globular, subcutaneous swelling of size 11 x 9cm over his left elbow for 1 year duration.(Figure 1) He had a history of trauma on his left elbow prior to the onset. He was neither a diabetic nor an immunocompromised individual. Systemic examination was normal. Aspiration cytology from the swelling revealed PAS positive branching septate fugal hyphae with brown melanin pigment, following which a diagnosis of subcutaneous phaeohyphomycosis was done. Pus was sent for fungal culture and sensitivity. On Sabouraud's Dextrose Agar, olivaceous black with slimy center due to yeast like growth and with floccose margin are seen on the obverse side with diffuse black pigmentation on the reverse.(Figure 2).Lactophenol cotton blue mount showed black pigmented septate hyphae with abundant budding yeast cells .Conidiophores appeared brown, cylindrical, thick walled with spine-like annellated apical region.Based on macroscopic and microscopic morphology correlated to that of *Exophiala spinifera,* a rare species to be isolated as an etiological agent. Surgical excision of the cyst with intact wall was done. HPE of the cyst wall revealed suppurative granuloma with fungal elements.

DISCUSSION

Phaeohyphomycosis is an infection caused by fungi that are characterized by a brown to black color within the cell wall of vegetative cells, conidia, or both. Dark pigmentation of the moulds is caused by the cell wall deposition of dihydroxynaphthalene melanin that is formed through pentaketide metabolism. Increased numbers of cases of phaeohyphomycosis have been observed in medical centers most commonly in large group of immunocompromised patient population[1].

Phaeohyphomycosis has a wide range of clinical presentations like superficial colonization of the skin, keratitis, subcutaneous cyst formation, allergic sinus disease, and fatal disseminated infections. Few studies have stated that relatively common presentation of phaeohyphomycosis is subcutaneous lesion, as that we have reported[3,4].

Clinical presentation of phaeohyphomycosis due to *Exophiala spinifera* **as single or few lesion in immunocompromised adults or disseminated** lesions in the immunocompetent has been reported in various part of India[5-8].

In our case, we had presentation of subcutaneous swelling in a immunocompetent individual for 1 year duration, whose was farmer by occupation. Infection occurs due to traumatic implantation of fungal material from contaminated plants and soil. Many cases of phaeohyphomycosis may have no evident exposure history[9].

On review of various the literature on phaeohyphomycosis, by Sharma *et al*[10], 23 patients with subcutaneous phaeohyphomycosis have been reported from India predominantly involving the extremities and very few cases with dissemination.

Our case report highlights the fact that any subcutaneous soft swellings must also be considered under differential diagnosis for fungal infections as they may be mistaken. Some of the differential diagnosis includes of this cyst includes ganglion cysts, epidermal inclusion cysts, Baker's cysts, or foreign body granulomas. Cysts in the immunocompetent patient presents usually as chronic, relatively asymptomatic and remain unnoticed, but a fungus in tissue can be observed and cultured when the cyst is removed. The most common dematiaceous fungi to produce these cysts are ***Exophiala jeanselmei, Wangiella dermatitidis, and Phialophora species.*** Among these, ***Exophiala jeanselmei*** is the major species producing clinical disease[3], whereas in our case we have identified ***Exophiala spinifera,*** which appears relatively very rare.

Diagnosis of phaeohyphomycosis relies on histopathology and culture. Identification of genera and species in the clinical laboratory is based almost exclusively on morphological study of macroscopic and microscopic features.

For a subcutaneous cyst, complete removal of the encapsulated structure can be curative but care must be taken not to leak contents into the wound. No serologic or non-culture-based methods are currently available to aid in the diagnosis of phaeohyphomycoses.

CONCLUSION

Although phaeohyphomycosis is a rare entity, there should be a high index of suspicion in cases with subcutaneous cystic swelling. This report highlights the potential role of *Exophiala spinifera* as an emerging cause of subcutaneous phaeohyphomycosis.

Figure 1: Subcutaneous swelling-Left Elbow Joint

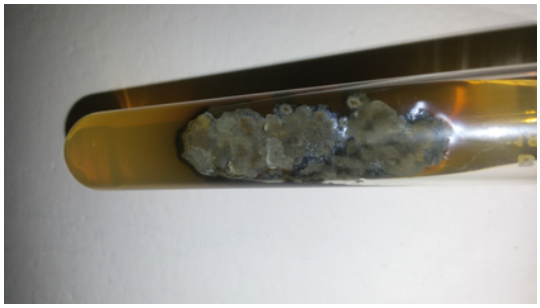

Figure 2: Sabouraud's Dextrose Agar showing growth of *Exophiala spinifera*

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