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Pathology

Subcutaneous Phaeohyphomycosis – A Rare Case Report

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Abstract **Case Report**

Phaeohyphomycosis is a heterogenous group of fungal infection caused by a variety of naturally pigmented fungi. A 56year old male presented with painless subcutaneous swelling over left anterior aspect of distal leg since 1 year. Local examination revealed a painless, fluctuant, soft, mobile swelling measuring 3x2 cm in size over left anterior aspect of distal leg. Microscopic examination revealed granulomatous inflammation with presence of septate fungal hyphae with constriction and occasional irregular branching. (Identical to Phaeohyphomycosis).

Keywords: Fungal infection, Pigmented fungi, Granulomatous inflammation.

INTRODUCTION

Phaeohyphomycosis is a subcutaneous or systemic infection caused by dematiaceous, mycelia-forming fungi [3]. Phaeomycotic cysts (Greek - dark) are superficial cystic granulomas caused by a fungus that is pigmented in tissue section. This entity is also known as phaeosporo trichosis, phaeohyphomycosis, hypodermomycosis, and subcutaneous abscesses caused by brown fungi [1]. This is a histopathologic definition of a disease process that can be caused by many different organisms and can have multiple different clinical presentations. Bipolaris, Phialophora, Alternaria, and Exophiala are fungi responsible for phaeohyphomycosis [4, 5].

Phialophora gougerotii is usually isolated from phaeohyphomycotic cysts, but other species have been recovered, and it is likely that many species of pigmented fungi can cause these lesions. Because the usual habitat of phialophorid fungi is on lumber, infection by traumatic implantation of plant material has been surmised, but clinical history of trauma or histopathologic evidence of vegetable matter have rarely been found [1]. Phaeohyphomycotic cysts are subcutaneous masses which evolve indolently with gradual central softening). The usual location is on the extremities, and deep muscular or bony involvement is rare. When a lesion is near a joint, the preoperative diagnosis may be synovial cyst of tendon sheath. In other locations, this lesion may be mistaken for lipoma or foreign body granuloma. (Figure 1) [1].

Figure 1 This mass on the dorsum of the foot of an African could be confused with a synovial cyst of tendon sheath

Most phaeohyphomycotic cysts are solitary and they do not provoke regional lymphadenopathy. Sinus tracts and epidermal involvement complicate phaeomycotic cysts only after aspiration. Immunosuppression may be a factor in some patients, but most lesions [12] studied at the AFIP have been from healthy people [1]. Phaeohyphomycotic cysts are encapsulated and shell out easily. Most are cystic, and the others have some central softening. The lining may be papillomatous, and the color of the pus varies from creamy white to tan [1]. These lesions have a characteristic pattern. The epidermis and dermis are usually normal. demonstrates a well circumscribed cyst lying in the subcutis. Its fibrotic wall encloses a layer of granulation tissue with sheets of epithelioid and giant cells. Centrally, focal collections of acute inflammatory cells produce microabscesses. Occasionally, the wall of the cyst contains foci of eosinophils.

The presence of yeast and hyphal forms of variably pigmented fungus establishes the diagnosis of phaeohyphomycotic cyst. Diligent search may identify an associated splinter in the tissue or liquid pus. The organisms are found [2]. The organisms are found cause them to resemble pseudohyphae or yeast forms, but true yeast forms are rare. Mycelia, if present, are more loosely arranged than the compact masses of hyphae seen in eumycetoma. Pigment is not always obvious and may be highlighted using theFontana-Masson stain [4]. The lesions have been cured by surgical extirpation [1].

Pathogenesis

The subcutaneous cystic type of phaeohyphomycosis is usually caused by Phialophora gougerotii (formerly called *Sporotrichum gougerotii*) [10]. Less commonly, it is due to Exophiala (Fonsecaea, Wangiella) dermatitidis [11]. Melanin is directly involved in the virulence of phaeohyphomycetes. Melanin is able to scavenge free radicals used by phagocytic cells to kill fungi, and melanin may also bind the hydrolytic enzymes used by phagocytic cells to lyse fungal cell membranes. Strains of these fungi that lack pigment demonstrate reduced virulence in mouse models [12,13]. Nondematiaceous strains have decreased resistance to fungicidal effects of the phagolysosome [14]. These factors may also help to explain the virulence of phaeohyphomycetes in immunocompetent patients.

CASE REPORT

A 56year old male patient presented with painless swelling over anterior aspect of left distal leg since 1 year duration. Initially the swelling was small but gradually increased in size. There was no specific history of local trauma. On examination, the general condition was good. Patient was afebrile. Other systemic examination results were within normal limits. Local examination revealed a painless, fluctuant, soft, mobile swelling measuring 3x2 cm. Over anterior aspect of left distal leg. It was not attached to underlying bone. Radiological investigations were not done. Pre surgical blood investigations were within normal limits. HIV and HBsAg were negative. The provisional clinical diagnosis was Inclusion dermoid cyst. Surgical excision of the swelling was done and sent for histopathological examination in 10% formalin.

Gross examination findings

Received a specimen of skin measuring 2.5x1.7 cm in size. Skin is ulcerated. Cut surface shows a cystic space measuring 1x0.3 cm. Divided and whole processed.

Microscopic examination findings

Sections shows a central absces cavity surrounded by a fibrotic cyst wall which shows chronic xanthomatous & chronic granulomatous inflammation along with multinucleated giant cells & necrosis. Grocott stain shows presence of septate fungal hyphae with constriction & occasional irregular branching. The findings are suggestive of subcutaneous phaeohyphomyosis (phaeohyphomycotic cyst).

Diagnosis: Subcutaneous phaeohyphomycosis, (phaeohyphomycotic cyst), left anterior distal leg.

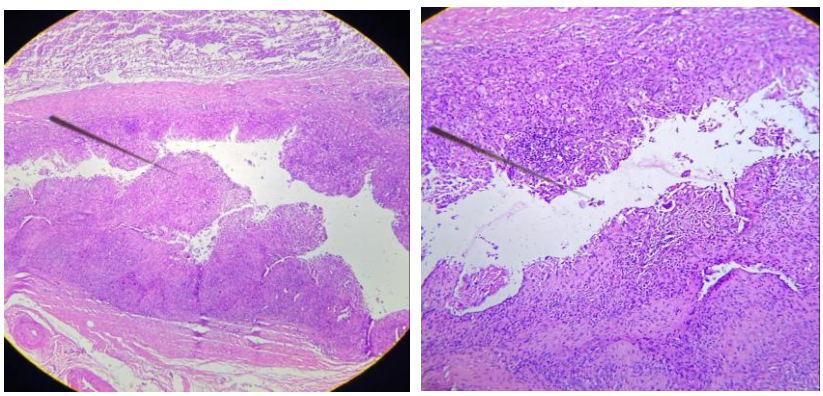

Figure 2: Chronic xanthomatous & chronic granulomatous inflammation along with multinucleated giant cells & necrosis. (10X & 40X)

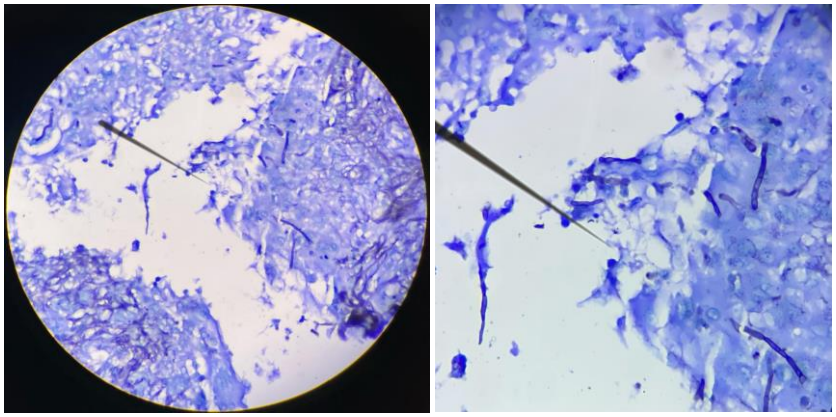

Figure 3: Grocott stain with presence of septate fungal hyphae with constriction & occasional irregular branching (10X & 40X)

DISCUSSION

Phaeomycotic cysts (Greek - dark) are superficial cystic granulomas caused by a fungus that is pigmented in tissue section. Others have called this entity phaeosporo trichosis, phaeohyphomycosis, hypodermomycosis, and subcutaneous abscesses caused by brown fungi [1].

Rare phaeomycotic cysts studied come from every continent except South America [1]. Because the usual habitat of phialophorid fungi is on lumber, infection by traumatic implantation of plant material has been surmised, but clinical history of trauma or histopathologic evidence of vegetable matter have rarely been found. Phaeohyphomycetes often infect people who are not overtly immunosuppressed. However, immunosuppression does increase the risk of infection with less commonly pathogenic phaeohyphomycetes such as *Alternaria infectoria* and increases the risk for disseminated disease [1].

Differential Diagnosis

The organisms of phaeohyphomycosis can be distinguished from Aspergillus sp. because the latter have hyphae with relatively uniform diameter and regular dichotomous branching [15]. Furthermore, disseminated aspergillosis is associated with vascular invasion, ischemic necrosis, and relatively less inflammation [2]. Regarding the management of subcutaneous phaeohyphomycosis, excision of the localised lesion is usually curative [16].

CONCLUSION

Subcutaneous phaeohyphomycosis is a rare fungal infection caused by a wide variety of dematiaceous fungi. A high index of suspicion for infective aetiologies is needed to make a proper clinical diagnosis. For a localised lesion, simple excision is usually curative. When phaeohyphomycosis is not suspected and the identification of species by culture is not pursued, routine histopathological examination is sufficient to arrive at a diagnosis of phaeohyphomycosis.

Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper

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