Cystic Subcutaneous Phaeohyphomycosis by Cladophialophora Species in an Old Leprosy Patient

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Abstract

A diverse collection of species known as phaeoid (dematiaceous) fungi are connected by the production of the pigment dihydroxynaphthalene melanin. Although they don't usually show up as human infections, phaeoid fungi are commonly found in nature as contaminants. Decomposing vegetation, rotting timber and soil all contain these fungi. On the other hand, a rise in infections is probably due to a rise in the population of individuals with compromised immune systems. An elderly female patient from southernmost part of India who was treated earlier for leprosy presented with multiple boggy swellings in right hand. Appropriate sample was collected under aseptic precautions and subjected for microbiological analysis where Cladophialophora species was isolated. The patient was managed with antifungal drugs but tilted towards poor outcome.

Categories: Pathology, Dermatology, Infectious Disease

Keywords: fluconazole, one celled blastoconidia, leprosy, cladophialophora, cystic subcutaneous phaeohyphomycosis

Introduction

Leprosy, sometimes referred to as Hansen's disease, is an infectious disease that is treatable but is still widespread in more than 140 nations worldwide [1]. In leprosy patients, the most commonly reported nonviral coinfections are Tuberculosis, Leishmaniasis, Chromoblastomycosis like infections and Helminths. Even though they are very much common, co-infections such as leprosy and chromoblastomycosis related infections are unusual [2]. Cystic Subcutaneous Phaeohyphomycosis is localized cutaneous infection following deep inoculation of fungus into subcutaneous tissues. The lesions are found on feet, legs, hands, arms, back or other body sites. The nodules develop as indolent, usually small subcutaneous masses [3]. In this report, we present the case of Cystic Subcutaneous Phaeohyphomycosis caused by Cladophialophora species in a diabetic woman who got recovered from Hansen's disease 50 years back. The pathogen was identified as Cladophialophora species by demonstrating characteristic colony morphology and microscopic features. Patient was treated with fluconazole but resulted in an unfavourable outcome. An increased awareness is necessary for appropriate treatment of such conditions. Cladophialophora species are highly neurotropic and invariably proven fatal hence frequently isolated from Brain abscess. This species has been involved in human skin lesions along with cases of chromoblastomycosis like infections and even it has recently been reported in a lung infection [3]. This article was previously presented as a poster at the SAI MICROCON 2023 INTERNATIONAL E-CONFERENCE on July 20, 2023.

Case Presentation

A 85 years old lady with agricultural background, a known diabetic on irregular treatment and recovered from Hansen's disease approximately 50 years back presented with complaints of multiple boggy swellings (Figure 1 and 2) in right hand, insidious onset progressive in nature for past two months. There was no history of numbness, sensory deficit in all four limbs, trauma or fever.





FIGURE 1: Multiple boggy swellings in right hand (Upper view)





FIGURE 2: Multiple boggy swellings in right hand (Side view)

From the patient, Pus aspirate sample was collected from boggy swelling and sent for fungal examination. Potassium hydroxide (KOH) mount revealed numerous irregular septate branched hyphae [3]. Sabouraud dextrose agar (SDA) with Chloramphenicol showed spread growth with olive grey to brown in colour (Figure *3*). Reverse is black (Figure *4*) [4].





FIGURE 3: Sabouraud dextrose agar with Chloramphenicol (Obverse)





FIGURE 4: Sabouraud dextrose agar with Chloramphenicol (Reverse)

Lactophenol cotton blue (LPCB) mount revealed one celled blastoconidia without evident darkly pigmented hila which are borne in long, sparsely branching conidial chains (Figure *5*) [3].





FIGURE 5: Lactophenol cotton blue (LPCB) mount

Periodic Acid-Schiff (PAS) stain revealed sheets of neutrophils admixed with eosinophils and Lymphocytes in necrotic background. Thin septate branched fungal hyphae seen (Figure 6 and 7).





FIGURE 6: Periodic Acid-Schiff (PAS) stain

Light green used as counterstain





FIGURE 7: Periodic Acid-Schiff (PAS) stain

Hematoxylin used as counterstain

Other relevant biochemical investigations were carried out which includes Complete blood count, Renal function tests, Liver function tests, Lipid profile, Thyroid profile, HbA1c as well as Chest X-Ray and Computed tomography (CT) scan of Brain. All reports were within normal limits. She was also nonreactive for Human immunodeficiency virus (HIV), Hepatitis B surface antigen (HBsAg), Antibodies to Hepatitis C virus (Anti-HCV tests). Based on the clinical picture and microbiological analysis, we diagnosed this case as Cystic Subcutaneous Phaeohyphomycosis. The isolate was presumptively identified as Cladophialophora species. Due to non-availability of effective medical treatment, early diagnosis and excision of lesion is indicated to avoid further complications [3]. Still, we proceeded for antifungal therapy after analysing the factors like age, immune status and comorbidities but results in poor prognosis and negative outcome.

Discussion

The suspicion of fungal infection arises whenever a patient with multiple boggy swellings along with comorbidities like diabetes mellitus and doesn't respond to the usual empirical therapy. Besides the well known phaeoid fungi e.g. Phaeoacremonium minimum, Exophiala jeanselmei, Pleurostomophora richardsiae, Exophiala salmonis are reported as human pathogens causing Cystic Subcutaneous Phaeohyphomycosis [5-8]. Successful outcome of Cladophialophora infection is related to underlying disease control and appropriate antifungal therapy like Liposomal Amphotericin B and azole derivatives like voriconazole [3].

Conclusions

Cladophialophora species has a predilection for central nervous system and consequently causes cerebral Phaeohyphomycosis. But now, there are some of the reports implicating this fungus as an agent of subcutaneous Phaeohyphomycosis and other systems like lungs. Thereby increasing knowledge about various species causes subcutaneous mycosis is very essential. Due to the emerging antifungal resistance effective preventive measures and knowledge about fungal species is urgently needed to curtail the infections.

Additional Information

Disclosures

Human subjects: Consent was obtained or waived by all participants in this study. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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