

Phaeohyphomycosis of the Face Masquerading as Basal Cell Carcinoma in an Immunocompetent Patient

Abstract

Phaeohyphomycosis is a rare heterogeneous group of mycotic infections caused by dermatiaceous (phaeoid) fungi affecting the skin, subcutaneous tissue, and the central nervous system. Involvement of the face is extremely rare, and very few cases have been reported in India so far. We report a case of phaeohyphomycosis in a 45-year-old female with 1-year history of a well-defined hypertrophic plaque over the right cheek advancing towards the forehead. The lesion was ulcerated with rolled-up margins; a provisional clinical diagnosis of basal cell carcinoma was given. Histopathology of the skin biopsy revealed numerous multinucleated giant cells and fungal hyphae with in and in between the giant cells. Various histochemical stains were used to confirm the presence of fungal hyphae. Melanin pigment in the fungus was demonstrated with Masson's Fontana stain. Based on the histopathological and histochemical findings, a diagnosis of phaeohyphomycosis was given and it was concluded that the disease was more of a histopathological than clinical diagnosis. This case is being reported due to its unusual presentation and it also highlights the importance of histopathology in the diagnosis of this rare disease.

Keywords: Masson's-Fontana, melanin, phaeohyphomycosis

Introduction

Phaeohyphomycosis, meaning dark fungus with hyphae, is a rare infection caused by dematiaceous/melanized fungi that are distributed worldwide. They can involve the skin, subcutis, paranasal sinuses, or the central nervous system.^[1] In the skin, these hyphae appear brown-walled due to their ability to deposit melanin pigment.^[2] Multiple cases of subcutaneous phaeohyphomycosis involving the leg, foot, toes, arm, left thumb, hand, nails, wrist, waist, or the buttock have been reported from India.^[3] Involvement of the face is extremely rare, and very few cases have been reported in India so far.^[1]

Case Report

A 45-year-old female from Nepal presented in the Dermatology outpatient department with 1-year history of a well-defined hypertrophic plaque over the right cheek advancing towards the forehead. The lesion showed ulceration and crusting with rolled-up margins. It was associated with itching and erythema along with atrophy and telangiectasia. She had no medical illnesses or any surgical procedures in the past. The

provisional clinical diagnosis of basal cell carcinoma (BCC) was considered, and a punch biopsy skin specimen from the forehead was taken for histopathology.

Hematoxylin and eosin-stained sections revealed epidermal hyperplasia and a diffuse, dense lymphohistiocytic infiltrate along with numerous foreign body and Langhans giant cells in the dermis. Branching, septate, brown-colored fungal hyphae were observed both within and in between the giant cells [Figure 1a and b].

Histochemical staining for periodic acid Schiff (PAS), Masson's-Fontana (MF) and Grocott's Methanamine silver (GMS) stains was performed. PAS stain revealed bright magenta-colored hyphae [Figure 2a], where as silver stain revealed brownish black hyphae with branching septae [Figure 2b]. MF demonstrated melanin pigment in the fungus [Figure 2c].

On histopathology, the final diagnosis of phaeohyphomycosis was made. Species identification could not be done as after the biopsy the patient did not return for the collection of her histopathological report and the specimen for culture could not be obtained.

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Sharma Shruti,
Avninder Singh,
V. Ramesh¹,
Fouzia Siraj

Department of Pathology,
National Institute of Pathology,
¹Department of Dermatology,
ICMR, Safdarjung Hospital
Campus, New Delhi, India

Address for correspondence:

Dr. Fouzia Siraj,
National Institute of Pathology,
ICMR, Safdarjung Hospital
Campus, New Delhi, India.
E-mail: fouziasiraj2009@gmail.
com

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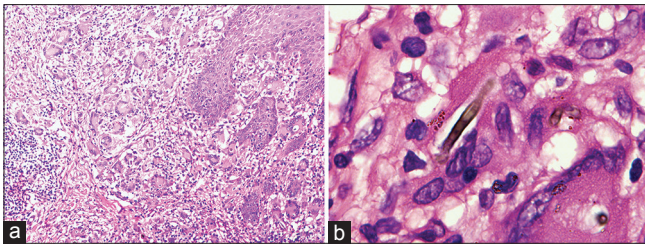


Figure 1: (a) Photomicrograph shows a dense lymphohistiocytic infiltrate along with numerous giant cells in the dermis. Brown colored fungal hyphae are seen both within and in between the giant cells (arrows). (Hematoxylin and eosin 10×). (b) High power view of the same showing brown colored septate fungal hyphae within the giant cells. (Hematoxylin and eosin 100×)

This case is being reported due to its unusual presentation and it also highlights the importance of skin biopsy and histochemical stains in the diagnosis of this rare disease.

Discussion

Phaeohyphomycosis is caused by a group of dermatiaceous fungi (brown moulds), which grow in tissues in the form of dark-walled septate mycelium. This mycelial tissue morphology separates phaeohyphomycosis from other types of brown-pigmented fungi where the tissue morphology of the organism is either a grain, as in mycotic mycetoma, or a sclerotic body, as in chromoblastomycosis.^[4] These opportunistic fungi mostly infect immunocompromised patients and may remain localized or spread via lymphatics.^[1,5]

Clinically, they present as superficial, cutaneous, subcutaneous, or systemic (disseminated) forms.^[6] Phaeohyphomycosis may range from a localized superficial infection of the stratum corneum, known as tinea nigra, to subcutaneous cysts known as phaeomycotic cyst to invasion of the brain.^[4] Subcutaneous phaeohyphomycosis is the most common sub-type and occurs due to wound contamination or traumatic inoculation of the saprophytic fungus from soil, decaying wood, and vegetation.^[7]

In our case, the lesion clinically mimicked BCC because it was ulcerated with rolled-up margins. A differential of lupus vulgaris was also considered keeping in view an ulcerated plaque-like presentation. Because on histopathological examination a definitive diagnosis of phaeohyphomycosis was made by the demonstration of dermatiaceous fungal elements in tissue, we can say that the disease is more of a histopathological than a clinical diagnosis. Phaeohyphomycosis is often seen in an immunocompromised state or preceded by a history of injury, however, both these predisposing factors were absent in our case.

The various agents causing phaeohyphomycosis are so similar in morphology that they cannot be differentiated solely on histopathology. Culture is the only definitive method of distinguishing various species of these fungi. Because these dermatiaceous fungi are well-known

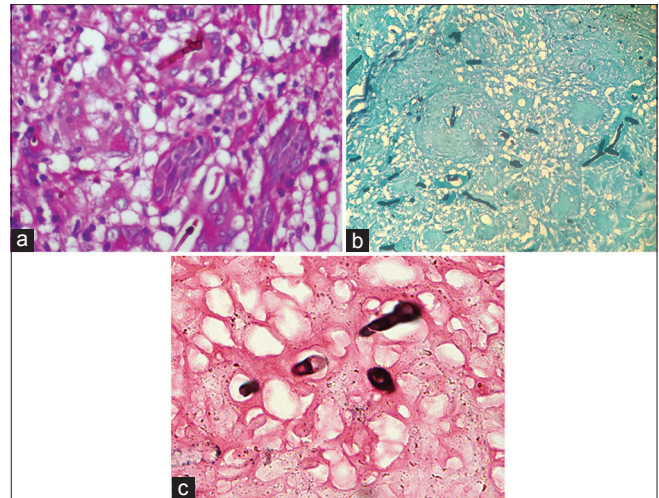


Figure 2: (a) Periodic acid Schiff (PAS) staining showing bright magenta colored fungal hyphae with branching and prominent septations. (PAS 40×). (b) Grocott's Methanamine silver (GMS) showing black-colored, septate, and irregularly branching fungal hyphae in the dermis. (GMS 40×). (c) Masson's-Fontana (MF) showing brown-to-black colored fungal hyphae in the dermis. (MF 100×)

airborne contaminants, a positive culture from a nonsterile specimen should be supported by direct microscopy to be considered significant.^[4] Hence, interpretation should be done in conjunction with the clinical presentation, culture report, direct microscopy, and histopathology.^[2] Various dermatiaceous hyphomycetes have been identified as the etiological agents, especially species of *Exophiala*, *Phialophora*, *Wangiella*, *Aureobasidium*, *Cladosporium*, *Curvularia*, and *Alternaria*.^[4] Of these, the most common is *Exophiala jeanselmei*.^[5]

In our case, culture could not be performed for the specific identification of the etiological agent as the patient was lost to follow-up. Identifying the specific agent is very important because different species may have tropism for different organs with varying response to antifungals. Gene sequencing of the internal transcribed spacer region is the most widely accepted molecular method because it can discriminate between inter and intraspecies variations.^[8]

Surgical excision is the chief modality of treatment with or without antifungal therapy. There are no standardized treatment protocols, however, itraconazole at a dose of 200 mg twice daily helps individuals who are unwilling to undergo extensive debulking surgery.^[9] While oral itraconazole is considered the empiric drug, voriconazole and posaconazole also show consistent *in vitro* activity against this group of fungi.^[8]

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Conflicts of interest

There are no conflicts of interest.

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