A rare case of phaeohyphomycosis caused by *Fonsecaea pedrosoi* in a child with nephrotic syndrome

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ABSTRACT

Dematiaceous fungi are the etiological agents of phaeohyphomycosis and are now increasingly being recognized for causing disease in humans. A high level of suspicion and routine fungal cultures are required to identify these cases. There is no consensus regarding their management. Here, an unusual presentation of phaeohyphomycosis (secondary to *Fonsecaea pedrosoi*) presenting as a disseminated infection in a case of nephrotic syndrome is described.

Key words: *Fonsecaea pedrosoi*, nephrotic syndrome, phaeohyphomycosis

Introduction

Phaeohyphomycosis is the term used to designate infections caused by demetaciaceous or pigmented filamentous fungi that contain melanin in their cell wall. During the last several decades, demetaciaceous fungi are increasingly being isolated from a variety of clinical conditions. Most cases of disseminated phaeohyphomycosis occur in immunosuppressed patients.[1] However, a few cases have been seen in immunocompetent individuals also.[2,3]

Nephrotic syndrome (NS) is one of the most common glomerular diseases affecting children. Based on the histological findings, minimal change nephrotic syndrome (MCNS) is seen in more than 80% of these children. Most patients with MCNS have favorable outcomes without complications. However, a few of these children are at a high risk for complications because of severe and prolonged proteinuria. The treatment of NS involves prolonged use of corticosteroids. Hence, complications arise not only due to the disease progression, but also as a consequence of the treatment.[4]

Here, we report an unusual presentation of phaeohyphomycosis secondary to *Fonsecaea pedrosoi* presenting as a disseminated infection in a child of NS.

Case Report

A 6-year-old girl with NS presented to the emergency at Kalawati Saran Children Hospital, New Delhi. The patient was diagnosed with NS at the age of 2 years when she presented to the emergency with the complaints of progressive swelling all over the body. Various investigations including blood tests, urinalysis, serum albumin, and imaging studies were done, and a diagnosis of NS was made. She was started on diuretics, corticosteroids, and albumin injections. She had improved but presented with similar complaints once a year for the last 4 years.

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In the present episode, she had complaints of facial swelling which progressed all over the body over a period of 1 week. She had fever for the last 3 days. The fever was intermittent and was associated with productive cough. She also had difficulty in breathing for the last 2 days. On examination, the patient had pitting edema all over the body, with a pulse rate of 78/min, blood pressure of 98/56, and a respiratory rate of 20/min. There were decreased breath sounds all over the chest, abdomen was tense, and tender with decreased bowel sounds. The patient was hospitalized and managed with adequate hydration, intravenous furosemide prednisolone at a dose of 2 mg/kg/day, and albumin infusion. Urinalysis showed 4+ protein, and a specific gravity of 1.030. Serum protein was 4 g/dl, serum albumin 1.8 g/dl, blood urea level of 68 mg/dl, and creatinine 2.6 mg/dl. Chest X-ray showed bilateral blunting of costophrenic angle suggestive of pleural effusion. A therapeutic pleural tap and ascitic tap was done and the samples were sent for bacteriological and fungal culture.

Direct examination of both the pleural and ascitic aspirate showed the presence of elongated septate pigmented hyphae. The aspirates were cultured on Sabouraud's Dextrose agar with gentamicin and chloramphenicol at 25°C, and after 2 weeks of incubation flat to dome-shaped colonies were seen to grow in both the aspirates. The colonies were dark olive gray to velvety with black reverse and developed radial grooves and a central elevation later. Slide culture revealed light brown septate hyphae and pale to brown conidiophores. Conidiogenous cells were arranged in loose branches showing sympodial arrangement with prominent denticles. Conidia were arranged singly and in short chains. The fungus was identified as *F. pedrosoi* [Figure 1].

On the basis of direct examination, the patient was started on intravenous liposomal amphotericin B at the dose of 5 mg/kg/day. She did not show any improvement and 1 week later, oral itraconazole was added at a dose of 2.5 mg/kg/day. The patient showed gradual improvement and was discharged on oral itraconazole for 4 weeks. Samples were repeated after a week which did not show any fungal elements.

Discussion

Phaeohyphomycosis is a clinical entity caused by dark-pigmented fungi. These demetaciaceous fungi more often cause skin and soft tissue infections, particularly due to trauma. However, systemic infections have also been documented. The etiological agents of phaeohyphomycosis can be numerous species belonging to different genera.[5]

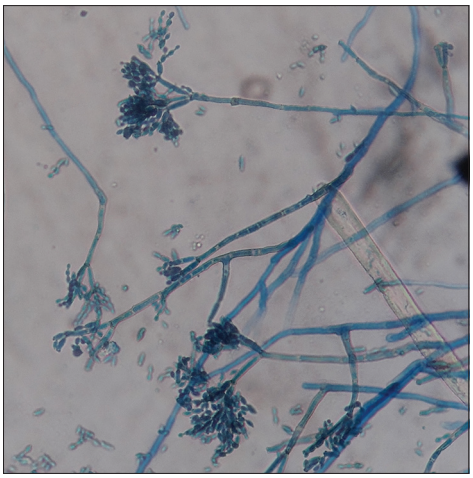

Figure 1: Lactophenol cotton blue mount of *Fonsecaea pedrosoi* showing thin septate hyphe and short chains of conidia

The commonest presentation of systemic phaeohyphomycosis is in the form of cerebral involvement like brain abscess, other localized infections such as pneumonia, pulmonary nodules, and endobronchial lesions.[2]

The most common isolates among demetacieous fungi are *Scedosporium apiospermum* causing disseminated infections, *Cladophialophora bantiana* in neurological, and *Ochroconis gallopava* showing pulmonary involvement.[2]

Fonsecaea is a pigmented, filamentous fungus found in rotten wood and soil. It is one of the major causative agents of chromoblastomycosis;[6] however, it is a rare cause of phaeohyphomycosis. A case of disseminated phaeohyphomycosis in a patient of renal transplant has been reported by Zaharapoulos at University of Texas Medical Branch, Galveston.[7] Two cases of cerebral involvement have been reported: Cerebral granuloma and cerebral phaeohyphomycosis.[3,6] *F. pedrosoi* has also been associated with corneal ulcer.[8,9] Mediastinal mass has also been reported by Singh *et al*. at PGIMER Chandigarh.[2]

The management of systemic phaeohyphomycosis requires extensive and timely medical intervention. It is a challenging condition to treat, mainly because of unresponsiveness to amphotericin B. There is no standardized therapy, and combination of amphotericin B with other antifungal agents, such as itraconazole, may improve survival rates as seen by Sharkey *et al*. and Rinaldi, who had concluded in two different studies that itraconazole could be used as the first-line therapy in mycosis, as it led to clinical stabilization and remission of disease.[10,11] In another study of the *in vitro* susceptibility by Bedout *et al*., on 12 primary human isolates of *F. pedrosoi*, resistance to amphotericin B, 5-flucytosine

and fluconazole was seen in 33.0%, 58.3%, and 66.7%, respectively, in contrast to 0% for itraconazole.[12] Itraconazole also proved to be effective in our case and the patient was cured. Voriconazole might have a role in therapy, but its role in clinical improvement is under study.[1]

F. pedrosoi is an important cause of phaeohyphomycosis besides being the most common etiological agent of chromoblastomycosis.

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Conflicts of interest
There are no conflicts of interest.

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