Case Report

Isolated frontal sinusitis due to *Pseudallescheria boydii*

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ABSTRACT

We report a case of recurrent sino-nasal fungal infection due to *Pseudallescheria boydii* (*P. boydii)* in a 33-year-old diabetic woman. It is very essential to identify *P. boydii,* as Miconazole is the only antifungal drug of choice. However, histological examination of the specimen for clinching clues such as intercalary conidia and chlamydoconidia plays an important role in identifying *P. boydii*, when fungal culture fails to yield the growth. On follow-up, the woman responded for the treatment with Miconazole and is free of symptoms, with no recurrence after 6 months.

KEY WORDS: Chlamydoconidia, intercalary conidia, Miconazole, *Pseudallescheria boydii*

INTRODUCTION

Isolated frontal sinusitis is rare, with disease due to *P. boydii* infection being even more unusual. *Aspergillus fumigatus* is the most common species implicated in paranasal sinus infection, with other species being rarely reported.[1] Like *Aspergillus*, *P. boydii* can colonize pre-existing cavities. Though less invasive than *Mucoracea* and *Aspergillus*, *P. boydii* can be invasive in severely immunosuppressed individuals.[2] To our knowledge, there has been only one previous reported case of *P. boydii* infection involving the frontal, maxillary and ethmoidal sinuses.[3] We believe this to be the first report of isolated frontal sinusitis due to *P. boydii* infection in a diabetic woman.

CASE HISTORY

A 33-year-old woman visited the department of Ear, Nose and Throat with complaints of recurrent throbbing headache since 2 years. She developed right side nasal blockade since 1 week. There was no history of trauma, watering of eyes and visual disturbances. She was a known diabetic and was on regular treatment since 10 years.

Nasal examination revealed post-septal deviation, middle meatal discharge, frontal floor tenderness and congested mucosa. Post-nasal discharge was observed during oropharyngeal examination. Her routine hematological investigations, urine examinations, blood sugar and urea were normal. She was found negative for HIV I and II by ELISA, tested two times 6 months apart.

In view of the history, a CT scan of her sinuses was arranged, which showed opacity of the right frontal sinus with erosion of frontal bone extending into superior wall of the right orbit [Figure 1]. A clinical diagnosis of recurrent frontal sinusitis with preseptal cellulitis was made and treated with ampicillin and gentamicin. Later, she underwent right middle meatal antrostomy and external exploration of frontal sinus with sinusectomy.

Histological examination revealed numerous grains in the lumen of the sinus tract [Figure 2A]. The grains revealed central pallor with peripheral eosinophilic fringe [Figure 2B]. The central pallor area showed clusters of intertwined, hyaline, refractile hyphal structures and irregular round-to-ovoid vesicular structures. There was cementing substance between the hyphae composing the grain. The Periodic acid-Schiff and Gomori's methenamine silver stained sections identified hyphal structures, which were narrow with large ovoid-to-round bulbous endings from hyphae (conidia or vesicles or chlamydoconidia), some of them showing branching (between 45 and 90 degrees) and irregular septations. Interestingly, bulbous swelling in between the septae, i.e., "intercalaryconidia", was seen among the majority of the septate hyphae, which gave the clue to the final diagnosis [Figure 2C]. Large accumulations of neutrophils in all stages of degeneration were seen to be clinging to the grain. The surrounding tissue contained a dense fibrosis and granulation tissue. The case was diagnosed as of pale grain mycetoma due to *P. boydii*. The patient was treated with Miconazole. On follow-up, the woman responded to the treatment and is free of symptoms, with no recurrence after 6 months.

DISCUSSION

P. boydii is a saprophyte of low pathogenicity frequently isolated from soil, manure and decaying vegetation, having a worldwide distribution, belonging to the group Hyalohyphomycosis.[4,5] *P. boydii* is also called *Allescheria boydii* or *Petriellidium boydii* or *Scedosporium apiospermum*. The usual history of *P. boydii* mycetoma is that of trauma or puncture wound to the feet,

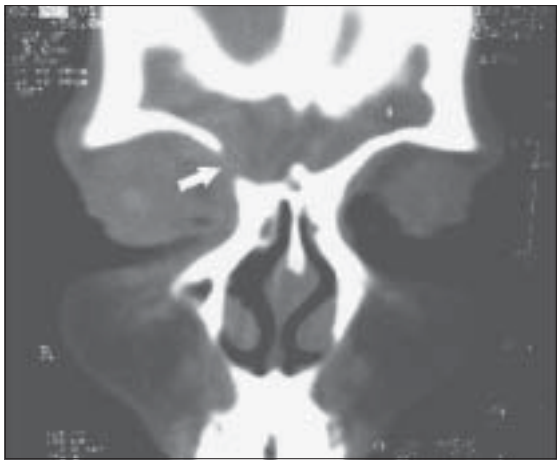

Figure 1: CT scan showing opacity of the right frontal sinus with erosion of frontal bone extending into superior wall of the right orbit

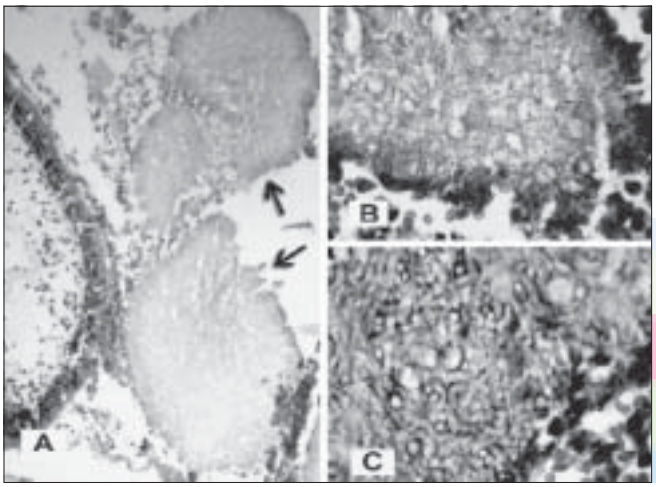

Figure 2: (A) (H&E, ×100) Histological examination showing numerous grains (arrows) in the lumen of the sinus tract. (B) (PAS, ×200) Grains with central pallor and peripheral eosinophilic fringe. (C) (GMS, ×400) The central pallor area shows clusters of intertwined, hyaline, refractile hyphal structures with chlamydoconidia and intercalaryconidia

legs, arms, or hands. *P. boydii* invades primarily subcutaneous tissues and ligaments; tendons, muscles and bones are usually spared. Sinuses and nasal septum may also act as a portal of entry for infections by *P. boydii*. Among the paranasal sinuses, the maxillary sinus is the commonest site involved, followed by sphenoidal sinus.

The age group affected is usually between 20 and 40 years of age. The occurrence is more frequent in men than in women (3:1 to 5:1). The usually associated predisposing factors are underlying disease, trauma, on immunosuppressive drugs, barrier breaks and also from aspiration of soil or swamp water or working in sewers.[6] Our patient was a 33-year-old diabetic woman; probably inhalation of soil dust might have led to infection.

In the present case, there was no growth on fungal culture. Failure to yield growth on fungal culture is not uncommon, may be attributable to overhomogenization of specimen or lack of viability of fungi.[7] Also, it is found that *P. boydii* was frequently isolated from a variety of saprophytic environment, but very rarely isolated from the clinical material.[4-6]

The grains of *P. boydii* on histopathology appear as pale eosinophilic, lighter central area with peripheral eosinophilic fringe. Surrounding the eosinophilic fringe are seen neutrophils and plasma cells. The pale central area of the grain shows dense mass of intermeshing hyphae.[8,9] The hyphae of *P. boydii* are very narrow with irregular septations and ends of these hyphae show chlamydoconidia. The diagnostic clue to identify hyphae of *P. boydii* is the bulbous swelling in between the septae (intercalaryconidia).[10] The above features seen in the present case were the clinching clues toward the diagnosis of *P. boydii*. Also, the patient responded to the treatment with Miconazole, which is the antifungal of choice for *P. boydii* infection.

P. boydii can produce potentially lethal complications in patients who are immunocompromised because of neutropenia, hematopoietic or solid malignancies, diabetes mellitus and acquired immunodeficiency syndrome. In these patients, almost any organ can be involved after the hematogenous dissemination of the fungus.[11] Hence, early diagnosis and management are crucial in preventing further complications.

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