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CASE REPORT

Cerebral phaeohyphomycosis: A rare case from central India

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ABSTRACT

Cerebral phaeohyphomycosis is a rare and frequently fatal disease, often caused by hematogenous spread of pathogens that are inoculated in the skin of the extremities following minor trauma, and its mortality rate is much high despite aggressive treatment. A rare case of 25-year-old immuno-competent man with cerebral abscess has been described. The etiological agent was microbiologically proved to be a dematiaceous fungi *Cladophilaophora bantiana*, which is primarily a neurotropic fungus causing deep-seated intra-cranial infections. Treatment was individualized by surgical intervention and appropriate anti-fungal therapy. Thus, it has been described as a successfully managed case of cerebral phaeohyphomycosis known to be associated with a high degree of morbidity and mortality.

Key words: Cerebral phaeohyphomycosis, *Cladophilophora bantiana*, dematiaceous fungi

Introduction

Cerebral phaeohyphomycosis is a rare and frequently fatal clinical syndrome diagnosed in patients with deep-seated cerebral infections secondary to dematiaceous fungi. Such fungi are common soil inhabitant, a true pathogen that are known for their neurotropism; however, central nervous system (CNS) seeding may occur through hematogenous route, probably initiated by respiratory colonization consequent to inhalation or through inoculation in the skin of extremities following a slight or minor trauma.[1] CNS phaeohyphomycosis presents with unusual features and is associated with poor prognosis without appropriate treatment.[2]

Ours is a rare case of cerebral phaeohyphomycosis from central India, presented by an immune-competent male patient with no history of trauma or sinusitis. It was managed successfully through surgical resection and anti-fungal therapy.

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Case Report

A 25-year-old unemployed male with no history of trauma or sinusitis presented with the complaints of severe bi-temporal headache accompanied with the fits of dizziness associated nausea, vomiting, and early papilledema. Examination showed a conscious and oriented young man who was able to communicate well but was clinically dehydrated (urea level raised to 55 mmol/L) His lumbar tap showed normal cerebrospinal fluid (CSF). Direct mycological examination of CSF was negative. His hematological and biochemical parameters were within normal limits except that he had a raised white blood cell count (15,900 cu/ml) with poly-morphnuclear leukocytosis (93%) and signs of microcytic hypochromic anemia. He was negative for human immunodeficiency virus test and hepatitis B surface antigen test.

Magnetic resonance imaging (MRI) scan of the brain showed a well-defined round to oval space occupying lesion with marked peri-lesional white matter edema in the right fronto-parietal lobe [Figure 1]. Postcontrast MRI brain axial image showed well-defined peripheral ring enhancement with mild lobulation along the inferior surface with no obvious solid enhancing component [Figure 2]. Thereby favoring the diagnosis of cerebral abscess with the least possible diagnosis of glioma. The patient was subjected to surgery. The brain specimen was multiple, irregular, friable grayish to grayish-white soft tissue mass bits of 2.5 cm × 2 cm × 1 cm that was sent for pathological investigation in the surgical pathology laboratory. The histopathological sections showed mixed inflammatory infiltration in the brain tissue with giant cells showing brown-colored, branched septate hyphae [Figures 3 and 4].

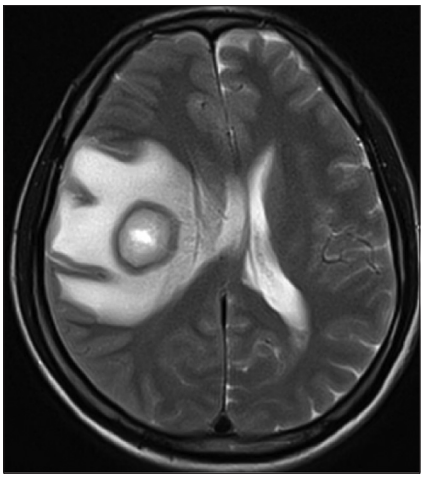

Figure 1: Magnetic resonance imaging scan of the brain showing a well-defined round to oval space occupying lesion with marked peri-lesional white matter edema in the right fronto-parietal lobe

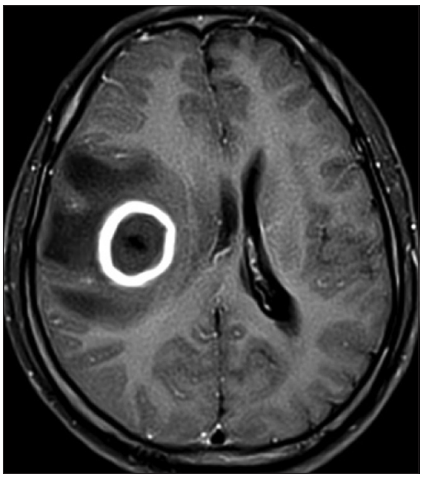

Figure 2: Postcontrast magnetic resonance imaging brain axial image showing a well-defined peripheral ring enhancement with mild lobulation along the inferior surface

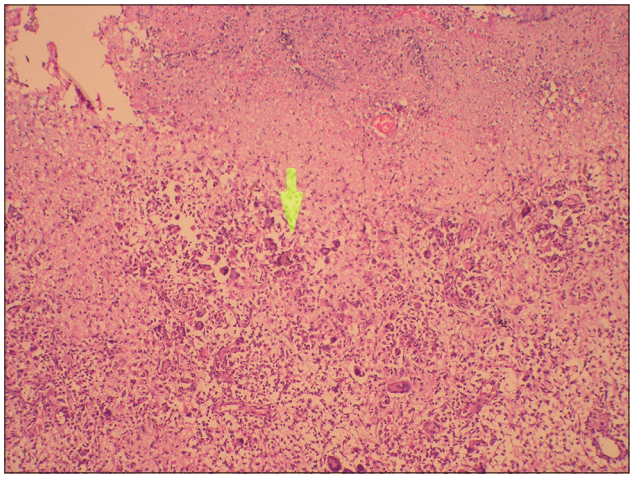

Figure 3: Mixed inflammatory infiltration in the brain tissue with giant cells showing brown-colored, branched septate hyphae (H and E, ×10)

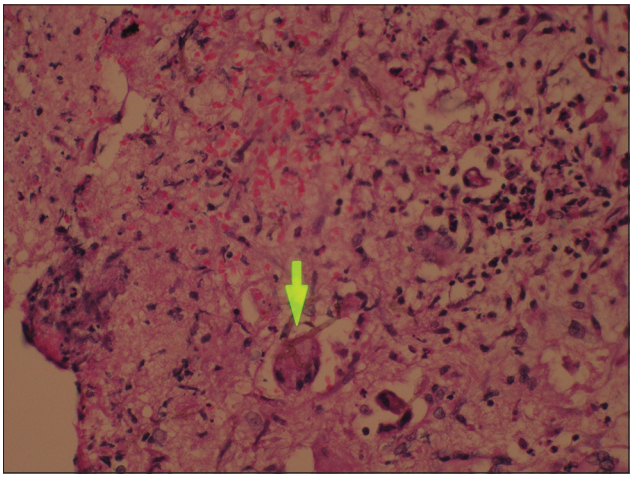

Figure 4: Mixed inflammatory infiltration in the brain tissue with giant cells showing brown-colored, branched septate hyphae (H and E, ×400)

Direct mycological examination of the cerebral tissue with 20% KOH showed the presence of elongated, branched, septate pigmented hyphae. Fragments of surgical brain specimen were cultured on 4% Sabouraud's dextrose agar (SDA) with chloramphenicol (50 mg/L). Incubation at 42°C revealed velvety, olive gray to brown growth on obverse and dark tan on the reverse of the SDA slant after 10 days of incubation. The etiological agent was identified as *Cladophilophora bantiana* (*Xylohypha bantiana*) and the final impression of cerebral phaeohyphomycosis was given.

The patient responded well to the surgical excision combined with antifungal therapy using itraconazole for 2 months. Unfortunately, the patient was lost for follow-up.

Discussion

Cerebral phaeohyphomycosis is a general term applicable to all brain infections where etiological agent is a dematiaceous fungi.[3] Out of the various species implicated in cerebral phaeohyphomycosis, most of them cause secondary infections following development of infection in other sites (mostly sinuses). Only four species like *C. bantiana, Exophiala dermatitidis*, *Ramichloridium mackenziei* and *Ochroconis gallopavum* are known to have a high neurotropic potential.[1,4] *C. bantiana* accounts for the majority of cases documented till date[5,6] and in our case also the etiological agent detected was *C. bantiana*. The fungus is known by several names such as *Cladosporium trichoides, X. bantiana, X. emmonsii* and *Cladosporium bantianum*. Approximately, more than 100 cases of cerebral phaeohyphomycosis caused due to different dematiaceous fungi have been documented in India out of which 28 cases are known to be caused by *C. bantiana*.[1,7,8]

Though the portal of entry is not well-established, inhalation of spores followed by colonization and subsequent hematogenous spread has been suggested.[2] Contiguous spread through

paranasal sinuses or trauma resulting into implantation of fungal elements into the skin and its subsequent dissemination may be the cause of disease.[5] However, in our case, the patient neither had sinusitis nor did he present any history of trauma, unlike the reports presented by few authors. Several studies have also correlated the cause of infection with the patient's occupation.[1,3,5] Though, the life-threatening fungal infections are often associated with severe immuno-compromised status, a typical but confounding and peculiar feature of this infection is that, it is increasingly recognized in immuno-competent persons with no underlying diseases.[1,2,9] The disease has also been frequently reported in males and in our case also the patient was an immuno-competent male.[1-3,5,9]

Histological examination of the causative organism is not enough to determine the type of fungus. Hence, second step must be to demonstrate the cultural aspects of the causative fungus.[8] In our case, we confirmed the etiological agent both through histological as well as microbiological investigations. Radiological investigations initially provided the extent of spread and later on the evidences of resolution so that the anti-fungal treatments could be individualized.[5] Treatments have largely been unsuccessful in most of the cases of cerebral phaeohyphomycosis. It must be individualized with surgical resection and optimized anti-fungal therapy. The azoles are commonly chosen in treatment strategies against cerebral phaeohyphomycosis since they exhibit excellent activity against dematiaceous fungi and can be prescribed safely for an extended period. Several authors have mentioned the use of amphotericin B in their treatment options, but itraconazole is even known to cure the relapses after therapy by amphotericin B.[2,4,9]

To conclude, cerebral phaeohyphomycosis should be considered in the differential diagnosis while examining the specimens from ring enhancing CNS lesions in immuno-competent patients especially males. Also, specimens should be handled with biosafety level of two containment because of the known pathogenesis of these organisms for immuno-competent individuals. The feasibility of surgical resection should be considered in all the patients of cerebral phaeohyphomycosis and duration of antifungal treatment should be individualized using radiological evidences of resolution.

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