Asian Journal of Pharmaceutical and Health Sciences
www.ajphs.com

Primary central nervous system phaeohyphomycosis: A rare case report

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ARTICLE HISTORY

Received: 17.09.2015

Accepted: 16.11.2015

Available online: 30.12.2015

Keywords:

cerebral phaeohyphomycosis, dematiaceous fungi. hyphae

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ABSTRACT

Phaeohyphomycosis are rare infections caused by dematiaceous fungi affecting the skin, subcutaneous tissue, paranasal sinuses or uncommonly the central nervous system, where it is associated with a high morbidity and mortality. Cladophialophora bantiana and Ramichloridium mackenziei, Xylohypha bantiana are the most frequent causes of cerebral phaeohyphomycosis; These fungi are extremely neurotropic and infections by them are usually confined to brain and meninges. Complete surgical removal associated with antifungal therapy is the treatment of choice. Early diagnosis is the key to proper management of this life threatening disease. We report a case of cerebral phaeohyphomycosis in an 11 year old child from Eastern India which was clinically and radiologically diagnosed as cerebral pyogenic abscess..

INTRODUCTION

Phaeohyphomycosis refers to infections caused by dematacious fungi which affect skin, subcutaneous tissue, paranasal sinuses and even brain.[1]These are opportunistic naturally pigmented fungi that are saprophytes of soil and wood. Infections can occur in healthy individuals but immuno compromised persons are more susceptible to develop infections of brain. Cerebral phaeohyphomycosis is rare and can be life threatening. The lesion usually presents as brain abscess and diagnosis can be made by demonstration of fungus either in cytology or biopsy. We present a rare case of primary cerebral phaeohyphomycosis in an 11 year old immunocompetent boy where diagnosis was made after histopathological examination of the supplied material from a suspected pyogenic brain abscess.

CASE REPORT

An 11 year old boy presented with headache and convulsions since 10 days. He was of average body built and non-diabetec, No cerebellar or meningeal signs were present. The patient was febrile, conscious, oriented and responded to verbal commands. All cranial nerve functions were normal. Systemic examination did not reveal any abnormality. Routine blood tests and cerebrospinal fluid examinations did not yield any diagnostic clue. Tests for HIV, HBS were negative.

CT scan of brain revealed a contrast enhancing ring lesion in the right parietal region. So a diagnosis of a cerebral pyogenic abscess was made. (Figure 1) Patient was operated and a cystic cavity with thick friable edematous wall containing pus was found. This was sent for histopathological examination. Gross received in Department of Pathology was multiple bits of greyish, white and irregular and friable tissue together measuring 2x1x1 cm.

Microscopic examination revealed a granulomatous lesion. Foreign body giant cells and a mixed inflammatory infiltrate were present. (Figure 2) Pigmented fungal elements present were engulfed by giant cells. (Figure 3) Fungi were black-brown with septate hyphae and yeast forms. So a differential diagnosis of Phaeohyphomycosis and Chromoblastomycosis was made, both of which are pigmented. Chromoblastomycosis was ruled out as morphologically they are modified yeasts seen as thick walled muriform cells or sclerotic bodies, present in small groups or collections. Where as phaeohyphomycosis is detected as small hyphae with septae showing constrictions and lightly brown in color. The patient was otherwise normal and did not have any evidence of fungal infections of skin or subcutaneous tissue. So a final diagnosis of primary cerebral phaeohyphomycosis was made and the patient was treated with surgical debridement and antifungals. He is under follow up and is reportedly doing well till date.

DISCUSSION

Fungal infections of the central nervous system have risen sharply due to acquired immunodeficiency syndrome,

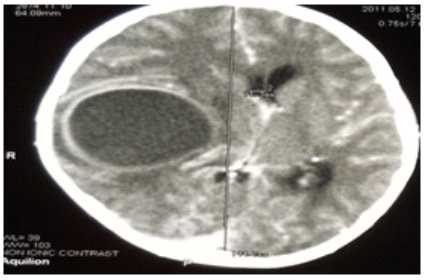

Fig 1 : CT scan showing contrast enhancing ring lesion in right parietal lobe

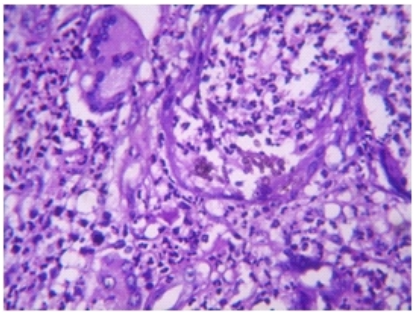

Fig 2 : Histopathology showing pigmented hyphae and yeast forms amidst mixed inflammatory infiltrate (H&E, X100)

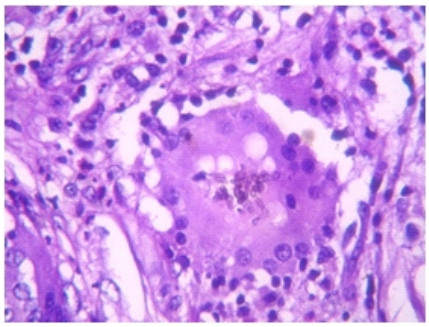

Fig 3 : Histopathology of fungus engulfed by foreign body giant cells (H&E, X400)

widespread use of broad spectrum antibiotics, steroids and immunosuppressive drugs. However, dematiaceous fungal infections of the central nervous system continue to be uncommon.[3] Phaeohyphomycosis is a rare infection caused by dematiaceous fungi affecting the skin, subcutis, paranasal sinus or central nervous system and are characterized by pigmented hyphae in the tissues.[1,5] Dematiaceous fungi have melanin-like pigment which make their recognition by H&E easy. It can also be demonstrated by special stains like Masson Fontana stain. The disease is a histopathological rather than a clinical entity.[1] This infection is endemic in the Middle East. In India, it has been reported from the extreme north to south, except from the western and eastern region. [1] We report this case from eastern India. Cerebral phaeohyphomycosis is rare and in this case has occurred without any predisposing factors like HIV. Cladophialophora bantiana is a neurotrophic fungus thought to be responsible. C.bantiana is the commonest dematiaceous fungus causing brain abscess. It affects immunocompetent hosts and individuals with no obvious predisposing factors , though fungal infections are usually associated with immunocompromised hosts.[2,3] Most infections occur in young males in the second and third decades of life. [3] Other fungi responsible for cerebral phaeohyphomycosis are *Ramichloridium mackenziei* and *Exophiala dermatitidis* The cerebral cortex is the commonest site, though the cerebellum , brainstem and spinal cord may also be involved.[4] About 101 cases of primary central nervous system phaeohyphomycosis were reported between 1966 to 2002. [5] CT scan reveals a unilateral well circumscribed mass lesion usually in the frontal lobe.[6] Diagnosis can be done from cytology of the lsion, KOH stain of the cytologic material, biopsy with special Stains and culture. In cerebral pheaohyphomycosis, the pathogen cannot always be cultured and identified from the serum and/or the CSF in which case biopsy of the lesion is recommended.[7] In histopathology, branching septate pigmented hyphae are found along with mixed inflammatory infiltrate and giant cells. Immunohistologic stain with direct fluorescent antibody can detect the organism. Serology and molecular tests are not of much help.

CONCLUSION

Although the prognosis of cerebral phaeohypho mycosis is grim, early diagnosis and prompt treatment may prove life saving. This case is being reported because of its rarity in this region and early diagnosis by histopathology.

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