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RESEARCH ARTICLE

PHAEOHYPHOMYCOSIS MENINGITIS IN AN IMMUNOCOMPETENT PERSON: A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Fungal meningitis in an immunocompetent host is generally uncommon, and infections due to brown-black or dematiaceous fungi are distinctly rare. These are distinct neurotropic organisms causing mostly cerebral abscess and paranasal fungal sinusitis. We report an unusual case of dematiaceous fungal meningitis in a man with no significant co-morbidities and no other identifiable risk factors for disease. CV junction meningeal biopsy was done which revealed this organism. Since these are so infrequently seen in clinical practice, randomized clinical trials are impractical and there is no consensus regarding treatment. He was treated with empirical antifungals and fortunately, he responded to treatment.

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INTRODUCTION

Central nervous system (CNS) infections caused by fungi are generally uncommon, and infections due to brown-black or dematiaceous fungi are distinctly rare. However, case reports are becoming more common and awareness of these infections is growing, in part due to widely publicized outbreaks of infection, often in association with contaminated therapeutics [Revankar, 2010; Smith et al., 2013; Centers for Disease Control and Prevention, 2002] Though many fungi have been implicated in disease, most cases are caused by only a few species, *Cladophialophora bantiana* being the most common. While there have been significant advances in the diagnosis of more common fungal infections due to *Candida* and *Aspergillus*, there are no specific diagnostic tests for dematiaceous fungi, adding to the challenge of their management. In addition, these infections are often refractory to standard drug therapies, and often require combined surgical and medical therapies [Mukherji, 1995]. Since these are so infrequently seen in clinical practice, randomized clinical trials are impractical and there is no consensus regarding treatment

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CASE REPORT

A 47 year old non-hypertensive, non-diabetic, euthyroid male, resident of Kolkata, West Bengal and teacher by profession presented to our hospital with a background history of fever, headache and neck stiffness for which he was diagnosed clinically as a case of tubercular meningitis. He was started on ATD (Isoniazid, Rifampicin, Pyrazinamide, Ethambutol) and steroids from May 2017. He developed ATD induced hepatitis and the regimen was rechallenged and pyrazinamide was replaced by moxifloxacin. Although his fever remitted but headache still persisted and repeat CSF revealed increasing protein and cell count. And MRI Brain revealed Enhancing nodular meningeal thickening / basal exudates with encasement of bilateral 3rd nerves, Right Optic nerve and Left Cerebellopontine angle nerves. He was continued on ATD and steroids reinitiated from July 2018. He presented to our side with a relapse of fever, headache and dimness of vision of Right eye, all for the past 3 months. The fever was low grade, intermittent with mainly evening rise. It was not associated with cough, rhinorrhoea, dysuria, pain abdomen. The headache increased in intensity and frequency and mainly localized to occipital and cervical region. The dimness in vision was gradual progressive, painless without any colour desaturation and affected both distant and near vision. There was no history of contact with TB, High risk behaviour, similar illness in his

Figure 1. CSF profile during the course of illness

family, visual problems, aura, tearing, photosensitivity, convulsions, altered sensorium, travel to endemic areas, exposure to cattles. His General examination did not reveal any obvious abnormality. His Bp was 120/70 mm Hg, Pulse rate 88 per minute and regular. His nervous system examination revealed a visual acuity of 6/18 in the Right eye and 6/9 in the left eye. There was no meningeal signs, no papilloedema, no other cranial neuropathy, no motor, sensory, cerebellar and autonomic abnormalities. Respiratory, cardiovascular and gastrointestinal system examination was essentially normal. Opthalmological examination revealed posterior subcapsular cataract probably due to chronic steroid use. Laboratory testing revealed mild leucocytosis of 11400/ul with monocytosis and ESR and CRP of 20 mm at first hour and 4.2 mg/dl respectively. His liver function test and renal function test was within normal limit and serology for HIV was non-reactive. His FBS was 96 mg/dl, PPBS was 324 mg/dl with an HBA_{1c} of 15.6 suggestive of steroid induced diabetes mellitus.

Analysis of the cerebrospinal fluid revealed an elevated protein level of 388 mg/dl (reference range, 15–45), glucose concentration of 216 mg/dl (reference range of 40–70 mg/dl) but white blood cells were increased at 1000 white cells/µl; 96% polymorphonuclear cells. CSF ADA, CBNAAT, Gram stain and culture, Indian ink preparation CSF Cryptococcal antigen and CSF VDRL were all non-contributory. CSF study also did not reveal any oligoclonal bands, and CSF IgG index was normal. His Chest X-ray, USG whole abdomen, ECG, Echocardiography were normal. MRI Brain revealed enhancing nodular meningeal thickening/basal exudates with enhancement and new lesions at CV junction, suggestive of Chronic basal meningitis. His Serum Lactate was 4.6 (reference <2mg/dl), S. ACE levels normal, ANA was negative, dsDNA, C3, C4 and IgG4 levels normal. Anti-Aquaporin 4 and Anti MOG were negative and VEP was normal. Finding no other alternative, meningeal biopsy was planned and biopsy from CV junction was done which revealed necrotising granulomatous lesion with numerous fragmented fungal hyphae which are slender pigmented and angulated by giant cells, Dematiaceous fungi. Thus, after almost two years of being treated with anti-tubercular drugs, a final diagnosis of chronic granulomatous meningitis with Dematiaceous Fungi with steroid induced diabetes mellitus and right posterior subcapsular cataract was made. ATD was finally stopped and an empirical antifungal therapy with Liposomal Amphotericin B and voriconazole was started. After 2 weeks, he developed amphotericin induced nephropathy and thus amphotericin was stopped but fortunately fever and headache remitted. Oral voriconazole and insulin was continued.

DISCUSSION

The diagnosis of phaeohyphomycosis can be difficult because dematiaceous fungi are commonly soil inhabitants and are often considered contaminants when identified in culture. No molecular techniques are presently available to rapidly and reliably identify these fungi to even the genus level. It requires strong clinical suspicion and invasive procedures for diagnosis. Most abundant fungal meningitis in our region is Cryptococcal meningitis, specially in the setting of an immunocompromised state. Dematiaceous fungi specially C. bantiana *is a rare cause of fungal meningitis. It mainly causes sinusitis and paranasal infections. But lately there have been increasing case reports of brain abscess and occasional meningitis caused by these organisms*. In a review by Revankar et al, C. bantiana accounted for 48% of 101 cases of primary CNS phaeohyphomycosis. Slightly more than half of the patients with CNS-limited disease had no underlying disease or risk factor for infection. There appears to be equal incidence between immunosuppressed and immunocompetent hosts, with male predominance. The pathogenesis of primary CNS phaeohyphomycosis is unknown at present. There have been reports of direct spread from paranasal sinuses. Another important aspect is the lack of definite treatment protocol and many regimens have been tried as we flip through the pages of literature. Amphotericin B, itraconazole, voriconazole have been used but no definite dose and treatment duration has been customized. Aggressive therapy is needed specially in cases of brain abscess.

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