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Apophysomyces Elegans A Cause of Maxillary Sinusitis - A Case Report

Dr Namrata Naithani, Dr Beena Hemanth

Department of Microbiology, M. S. Ramaiah Medical College and Teaching Hospital, Bengaluru, Karnataka, India

Correspondence Author: Dr Namrata Naithani, Department of Microbiology, M. S. Ramaiah Medical College and Teaching Hospital, Bengaluru, Karnataka, India

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Abstract

Mucormycosis is a rare necrotizing infection caused by a fungi within the class *Zygomycetes* and the order *Mucorals.. These affect immunocompromised as well as,* the genera *Apophysomyces* and *Saksenaea* can initiate invasive disease in apparently normal hosts.

Here, we report a rare case of mucormycosis in a 45-year-old male patient presenting with left sided nasal discharge with headache. Aspirated material from the left maxillary sinus was examined and was suggestive of Mucormycosis. The fungi on culture was identified as *Apophysomyces elegans*. The patient was started on liposomal Amphotericin B and underwent fenestrated Endoscopic sinus surgery with septoplasty and synechiae release ,showing an improvement.

Introduction

Mucormycosis is a rare necrotizing infection caused by a fungi within the class *Zygomycetes* and the order *Mucorals. This has emerged as an important fungal infection associated with high mortality rates especially in the last two decades.*[(1)]

The genera reported to cause invasive infection include Absidia, Mucor, Rhizomucor, Rhizopus, Apophys omyces, Saksenaea, Cunninghamella, Cokeromyces, and Syncephalastrum, with the first four being the most commonly reported pathogens.[(2)]

Based on anatomic localization, mucormycosis can be classified as one of the 6 forms: (1) cutaneous,(2) rhinocerebral,(3) pulmonary (4) gastrointestinal, (5) disseminated, and (6) uncommon presentations. Invasive mucormycosis has most commonly been reported from sites such as sinuses (39%), lungs (24%), and skin (19%). 23% of these cases later developed disseminated infections.[(3,4)]

Though mucormycosis infection is mainly seen in immunocompromised hosts, the genera *Apophysomyces* and *Saksenaea* can initiate invasive disease in apparently normal hosts who have sustained penetrating trauma during accidents.[(4)]

A. elegans was first isolated in India. Misra et al [(5)] in 1979 demonstrated this new fungus in soil samples collected from a mango orchard. *Apophysomyces elegans* is known to cause cutaneous, subcutaneous, and soft tissue infections following trauma, burns, or invasive procedures in apparently healthy individuals.[(6)]

Case Report

A 45 years old male patient came with a complaint of left sided nasal discharge for the last 1 year. The discharge was associated with headache. He was a known smoker and a non -diabetic. He gave a history of fenestrated sinus surgery done 5 years back. There was no history of

allergic rhinitis or recurrent rhinitis. There was no history of epistaxis , fever or any known allergies.

On examination , it was observed that the patient had synechiae in the left nasal cavity along with left sided deviated nasal septum which had resulted in decreased air entry in the left sided nostril.

Aspirated material from the maxillary sinus was sent for Potassium hydroxide mount (KOH Mount)which showed the presence fungal elements. A Nasal swab was also sent for aerobic culture and sensitivity which yielded no growth. A tissue histopathology specimen was sent from the left maxillary sinus which showed inflammatory polyp with fungal infection which was suggestive of Mucormycosis.

The specimen was inoculated on solid and liquid media such as Blood agar (BA), Thioglycollate media, Sabouraud's dextrose agar (SDA) plate with and without antibiotics, and incubated at 37°C and 25°C respectively. Within 3 days, macroscopically fast-growing, creamy white, and cottony colonies were seen. The reverse was white to pale yellow. (Fig 1)

A direct tease mount as well as slide culture revealed broad hyaline aseptate hyphae without any sporulation.

To induce sporulation ,the agar block method described by Ellis and Ajello (1982) was done.

A small block of agar was cut from a well established culture grown on PDA and was placed in the centre of petri dish containing 1% agar in distilled water. After 21 days at 26°C sporangium formation at the periphery of the petri dish was observed.(Fig 2)

Sporangiophores with distinctive funnel shaped apophyses and hemispherical-shaped columellae, with a conspicuous pigmented sub-apical thickening which constricted the lumen of the sporangiophore below the apophysis was seen .The fungi was identified as *Apophysomyces elegans*.(Fig2)

The blood investigation revealed the following values: Hemoglobin-15.2 g/dL, total leucocyte count-7880/mm3, platelet count- 1.5 lakhs/cumm ,prothrombin time-10.9 seconds, Serum aspartate aminotransferase-24 IU/mL(normal-30 IU/mL), alanine transaminase- 42 IU/mL (normal-30 IU/mL), random blood sugar-110 mg/dL serum creatinine-0.95 mg/dL ,sodium-143 mEq/L , and potassium- 5.16 mEq/L Patient was non-reactive for Human Immunodeficiency virus, Hepatitis C virus and Hepatitis B surface antigen.

No abnormality was seen in central nervous system, respiratory system or cardiovascular system.

The patient was treated with liposomal Amphotericin B and showed a good response. He again underwent a fenestrated Endoscopic sinus surgery with septoplasty and synechiae release, showing an improvement.

Figure 1: Culture Of Apophysomyces Elegans on Sabouraud's Dextrose Agar.

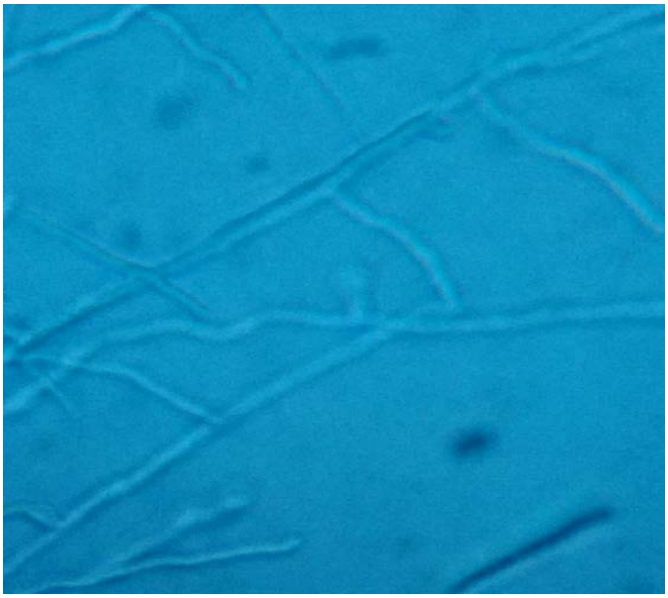

FIGURE 2- LPCB Mount from 1 % water Agar method showing bell shaped Sporangiophores.

Discussion and Conclusions

Mucormycosis is an emerging angioinvasive infection caused by the filamentous fungi of the class Zygomycetes.[1]The major predisposing factors for Mucormycosis to develop are hyperglycemia, ketoacidosis, corticosteroid therapy, malignancy, leukopenia,and immunosuppressive drugs.[7]

Zygomycetes can also cause devastating disease in persons with no underlying condition. Males are more commonly affected as compared to females.[6]

Aspergillus, Mucor, Rhizopus, Absidia are a more common causes of sinusitis.

Apophysomyces elegans is an emerging zygomycete that has been reported to cause invasive cutaneous and rhino-orbitocerebral infections in immunocompetent individuals as well. Once considered a rare pathogen, *Apophysomyces elegans* has increasingly been isolated from patients in tropical and subtropical climates over the last 2 decades. *A. Elegans* infection cases have been documented from India,southern United States, Australia, Mexico, Caribbean islands,Colombia, and Venezuela.[3,4]

Due to a rise in the cases of zygomycosis after traumatic inoculation as a result of accidents or natural disasters, *Apophysomyces elegans* seems to have become an emerging pathogen, especially in the Indian subcontinent.[4]

Infection by Apophysomyces species differs from other zygomycetes infections in two ways. Firstly, it occurs more frequently in immunocompetent than immunocompromised individuals, whereas other zygomycetes primarily infect hosts with weakened immune systems. Secondly, Apophysomyces infections are acquired directly by traumatic implantation rather than by inhalation of spores which then progress to rhino-cerebral or other disseminated infection.

The most common site of disease manifestation of *A. elegans* infection is the cutaneous and subcutaneous tissue, with local invasion into muscle and fat tissue resulting in necrotizing fasciitis. Thrombosis with extensive necrosis of the involved tissues is common.

Aggressive management including surgical debridement with liposomal Amphotericin B therapy is required until follow up evaluations show no residual fungal infections.[7]As Apohysomyces elegans is not a usual cause of sinusitis, hence we present an uncommon case report.

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