

Case Report***Apophysomyces elegans* causing Rhino-orbito Mucormycosis**Akhuji¹, A Banerjee¹, S Roy¹, K Karak¹, B Saha Dalal¹, S Bhattacharya¹*Sri Lankan Journal of Infectious Diseases 2014 Vol. 4 (1):51-54*DOI: <http://dx.doi.org/10.4038/sljid.v4i1.5861>Key words: *Apophysomyces elegans*; rhino-orbito mucormycosis**Abstract**

We describe here is a recent case of mucormycosis caused by *Apophysomyces elegans* in a 60 year old female from South Bengal with uncontrolled diabetes mellitus for two years. She presented with massive orbital oedema and pain in the periorbital area with proptosis of the right eye. Deep tissue obtained by endoscopic sinus sampling from the right middle meatus grew *Apophysomyces elegans*. The diagnosis was confirmed by histopathology which showed aseptate broad hyphae. Computed tomography scan showed right sided pan-sinusitis involving the right maxillary, ethmoid, frontal and sphenoid sinuses with an antrochoanal polyp and right retro orbital involvement of soft tissue. She required eye exenteration and extensive surgical debridement in addition to intravenous amphotericin B and antidiabetic drugs. She responded to the treatment.

Introduction

Mucormycosis is an infection caused by fungi classified within the class Zygomycetes. The genera reported to cause invasive infection include *Absidia*, *Mucor*, *Rhizomucor*, *Rhizopus*, *Apophysomyces*, *Saksenaia*, *Cunninghamella*, *Cokeromyces*, and *Syncephalastrum*, with the first four being the most commonly reported.¹ These fungi can produce serious and rapidly fatal infections, especially in the immunocompromised patient. *Apophysomyces elegans* has been reported as an emerging pathogen that can cause primary cutaneous zygomycosis in healthy patients.² Inhalation is the natural route of infection in rhino-orbito-cerebral infections.³ However, traumatic inoculation has also been described, particularly with *Apophysomyces elegans*.¹

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Case report

A 60 year old female from South Bengal with uncontrolled diabetes mellitus of two years, presented with a history of swelling and pain of the right eye of 20 days duration. Her pain worsened and periorbital swelling increased over the subsequent week. She also complained of progressive weakness and lethargy. On examination, her right eye showed massive cellulitis involving the eye lids, conjunctival congestion and ciliary congestion with a sluggishly reacting pupil. The anterior chamber was shallow and opacifications were seen in the lens. Examination of the left eye appeared normal throughout the period of admission.

On admission to the hospital, her haemoglobin was 9.1gm/dl, total leucocyte count was 14,900/cumm with a platelet count of 130,000/cumm. Her plasma glucose was 350g/dl whilst the serum creatinine and blood urea were 1.2mg/l and 52mg/dl respectively. Total serum protein was 6.0g/l with albumin of 3.9g/l and globulin 2.1g/l. A computed tomography (CT) scan of the head, orbit and paranasal sinuses showed retro-orbital soft tissue involvement suggestive of right orbital cellulitis and mucosal thickening in the right maxillary, ethmoid, frontal and sphenoid sinuses. Polypoid mucosal thickening was also noted in the right nasal cavity extending into the right side of the nasopharynx, suggesting right sided pansinusitis with an antrochoanal polyp.

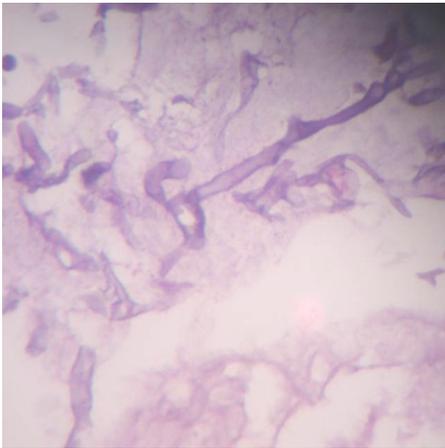


Figure 1:
Haematoxylin & Eosin stain shows broad sparsely septate hyphae (40X)

Endoscopic biopsy of the right maxillary sinus was carried out and sent for histopathology and microbiological examination. On histopathological examination, broad sparsely septate hyphae were seen embedded within the inflamed stromal tissue along with scattered dead membranous bones and some seromucinous glands (Figure 1).

The biopsy sample was inoculated onto Blood agar, MacConkey agar and Sabouraud dextrose agar (SDA) and incubated at 37°C. A fluffy, cottony textured colony with abundant aerial hyphae was grown on SDA after 48 hours incubation while no growth was observed on the other culture media. This white colony turned to brownish colour with age and no reverse pigment was noted (Figure 2).

Lactophenol cotton-blue mount showed broad sparsely septate branched hyphae suggestive of Zygomycetes but without sporulation. Distilled water yeast extract medium was inoculated and incubated at 37°C to obtain spores. After ten days



Figure 2:
Sabouraud Dextrose Agar with culture of *Apophysomyces elegans*

of incubation, microscopic findings showed sporangiophores with funnel-shaped apophyses and pyriform sporangia (Figure 3). Hence, this isolate was morphologically identified as *Apophysomyces elegans*.

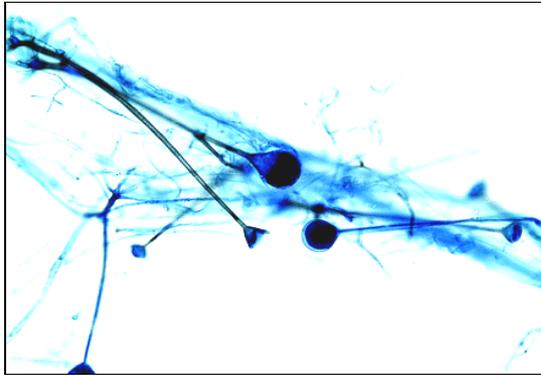


Figure 3:
Lacto-phenol cotton blue wet mount
showing distinctive funnel shaped
apophyses

Eye exenteration and extensive surgical debridement was carried out. The patient was treated with the antifungal agent, deoxycholate amphotericin B. Belonging to a poor socioeconomic strata, she could not afford liposomal preparation of amphotericin nor newer treatment options like Posaconazole. Due to frequent derangement in renal function tests and hypokalemia she only received a total dose of 21mg/kg of amphotericin deoxycholate. Her diabetes was managed with antidiabetic drugs. She responded well to the treatment.

Discussion

Apophysomyces elegans resembles *Saksenaea vasiformis* in gross colony morphology and its failure to sporulate on routinely used media.² *Apophysomyces elegans* has certain morphological features similar to those of *Absidia* species like the sporangiophores arising typically internodally and not opposite to rhizoids. *Apophysomyces elegans* shows the presence of hyphal foot cells similar to *Aspergillus* species.⁴ However, the presence of sporangiophores having funnel-shaped apophyses and pyriform sporangia differentiates *Apophysomyces elegans* from others.⁵

Liang *et al.* (2006) have reviewed 7 cases of rhino-orbito-cerebral mucormycosis caused by *Apophysomyces elegans*.¹ Two case reports of rhino-orbito-cerebral mucormycosis caused by *Apophysomyces elegans* have also been reported by Chakrabarti A *et al.* (2003) from India.³ Our case is similar to the case report described by Liang *et al.* (2006) including the symptoms, signs, treatment and outcome. *Apophysomyces elegans* shows good response to treatment with amphotericin B.⁶ Seventy one percent of patients with mucormycosis showed complete or partial response when treated with amphotericin B lipid complex.⁷ In our case, the patient responded to the treatment with amphotericin B along with surgical intervention and antidiabetic drugs. She is now on follow up.

Recommendations

Apophysomyces elegans is increasingly being reported as a cause of rhino-orbito-cerebral zygomycosis. The pattern of infection varies from mild infection to acute fatal infection. In most cases, *Apophysomyces elegans* has been reported to have favourable outcome after early diagnosis and treatment. On histological examination the morphology of *Apophysomyces elegans* is similar to that of other species of zygomycetes.⁸ Therefore, the possibility of *Apophysomyces elegans* infection must be considered in the differential diagnosis. The simple procedure by Padhye *et al.* (1988) of using distilled water yeast extract medium for sporulation can be practiced for identifying all nonsporulating zygomycetes isolated from clinical specimens in

routine laboratories.⁵ The specific and rapid identification of *Apophysomyces elegans* from clinical specimens, especially for all non-sporulating zygomycetes, along with histopathological findings is important because of the rapidity with which they invade the tissues and moreover to start treatment with antifungals at the earliest.

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