Apophysomyces elegans, a cause of malignant otitis externa complicated by skull-base osteomyelitis in an otherwise healthy non-diabetic individual: an uncommon form of mucormycosis from North India

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Background: Fungal Malignant otitis externa (MOE) leads to serious morbidity in elderly patients with diabetes mellitus. Over the last decade, mucormycosis caused by A. elegans has emerged as an important disease affecting immunocompetent individuals. We present a successfully treated case of an uncommon form of mucormycosis with A. elegans, causing fungal MOE complicated by skull base osteomyelitis, in an otherwise healthy non-diabetic individual. Clinical Case: A previously healthy 35-years-old non-diabetic female was admitted in Neuro-otology ward with 4-months history of severe left sided earache, ear discharge, decreased hearing and left sided multiple cranial nerve palsies, subsequent to an episode of left ear canal cleaning from a local quack by a wooden-stick. Patient's vitals, other clinical and biochemical parameters were within normal limits. Later, she developed multiple abscesses in posterior triangle neck and forehead on left side. MRI Brain revealed skull base osteomyelitis with diffuse patchy meningeal involvement. Aspirate from abscesses was sent for histopathological examination (HPE) and microbiology laboratories. HPE showed broad aseptate hyphae with right angle branching. No findings were suggestive of malignancy, tuberculosis & bacterial infection. Liposomal Amphotericin B (1mg/kg/day IV) was started and patient underwent an extensive surgical debridement and craniectomy of frontal bone.

Methods: Frontal bone tissue specimen after surgical debridement, was cut into small pieces for direct KOH mount microscopy and cultured on Sabauraud’s Dextrose Agar (SDA) (25°C, 37°C).

Results: KOH wet mount of the frontal bone tissue showed a very few broad, aseptate, thin hyaline hyphae with irregular non-dichotomous branching characteristic of Mucormycetes. Fungal culture of the frontal bone tissue on SDA (37°C) yielded scanty growth of white cottony fluffy colonies with yellow pigmentation after 7 weeks of incubation without any sporulation. Hence, sporulation was induced using water agar culture technique (37°C). Lactophenol-Cotton blue wet mount examinations revealed hyphae with rhizoids, sporangiophores, with a conspicuous, dark pigmented thickening below the funnel or champagne glass shaped apophysis & smooth walled subglobose sporangiospores. Phenotypically, it as identified as Apophysomyces elegans. Postoperatively, patient was started with combination therapy of liposomal Amphotericin B (1.5mg/kg/day IV) and Posaconazole (600 mg/day orally). The patient showed clinical improvement with liposomal Amphotericin B (cumulative dose:
2.5g; 3 months) and Posaconazole (for 6 months). Follow-up clinical examinations up to six months showed no evidence of persistent infection till date.

Conclusions: *A. elegans* can cause complicated fungal MOE, following a trivial skin trauma with contaminated wooden stick while ear cleaning in an otherwise healthy non-diabetic individual. Water agar culture is a simple method for inducing sporulation in *Apophysomyces* and rapid fungal identification in resource poor settings. High index of clinical suspicion, prompt treatment with extensive surgical debridement and Amphotericin B formulations along with Posaconazole are of great significance to survival outcomes of mucormycosis.