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Case Report

Orbital apex syndrome due to scopulariopsis: a rare case report

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ABSTRACT

Orbital infection with fungi of the order Mucorales, termed mucormycosis or zygomycosis; is sometimes seen in immunosuppressed patients, including those with diabetic ketoacidosis and malignancy. Presentations of scopulariopsis usually mimic that of mucormycosis, diagnosis is also difficult as initial smear on potassium hydroxide mount shows a similar picture to mucormycois. We describe a case orbital infection caused by Scopulariopsis in a patient with uncontrolled diabetes who presented with acute onset of unilateral painful ophthalmoplegia, defective vision and drooping of eyelid. Surgical debridement followed by Amphotericin B and itraconazole treatment responded as well.

Keywords: Scopulariopsis, Orbital apex syndrome, Invasive fungal sinusitis, Amphotericin B, Ophthalmoplegia

INTRODUCTION

Scopulariopsis are filamentous fungus found on decaying vegetation and in soil. Classified among the saprophytic hyaline hyphomycetes, these organisms grow readily at room temperature. The word scopula is Greek for broom, which describes the striking shape of Scopulariopsis conidiophores.[1] The presence of abundant conidia causes the colonies to appear yellowish-brown, with a powdery surface.[2] These organisms are hyaline (glassy) molds with septate hyphae. Scopulariopsis species are rarely considered pathogenic. Most commonly, they are implicated in onychomycosis.[3] There have also been reports of otomycosis, keratitis, prosthetic valve endocarditis, sinusitis, brain abscess, and cutaneous, subcutaneous and bone invasion by these pathogens in both immunocompetent and immunosuppressed individuals.[4]

CASE REPORT

A 63 year old woman presented with right sided headache, drooping of right eyelid and diplopia of one week duration. The patient is a known diabetic and hypertensive, was not taking any medication for the past four months. On examination, there was total ophthalmoplegia and complete ptosis. Corneal sensation was normal. Right eye pupil was 5 mm showed sluggish reaction to both direct and consensual reflex. Left eye pupil was brisk on direct and sluggish on consensual reflex. Vision of right eye was counting fingers close to face and left eye was 6/36. At the time of admission her blood pressure was 160/100 mmHg, fasting blood glucose was 277mg% and post prandial was 344 mg%. Urine ketone bodies were negative. Computerized tomography scan showed inflammatory mucosal thickening of sphenoid, maxillary, ethmoid and frontal sinuses. Magnetic resonance imaging showed significant mucosal disease in the right sphenoid sinus resulting in mild lateral bulge into the right cavernous sinus region corresponding to anatomical course of 3rd, 4th, 5th and 6th cranial nerves (Figure 1 A and B). The patient underwent nasal endoscopy under local anesthesia; right sphenoid ostium was identified and widened, revealed a polypoidal mucosal thickening with no evidence of bone erosion in sphenoid sinus.

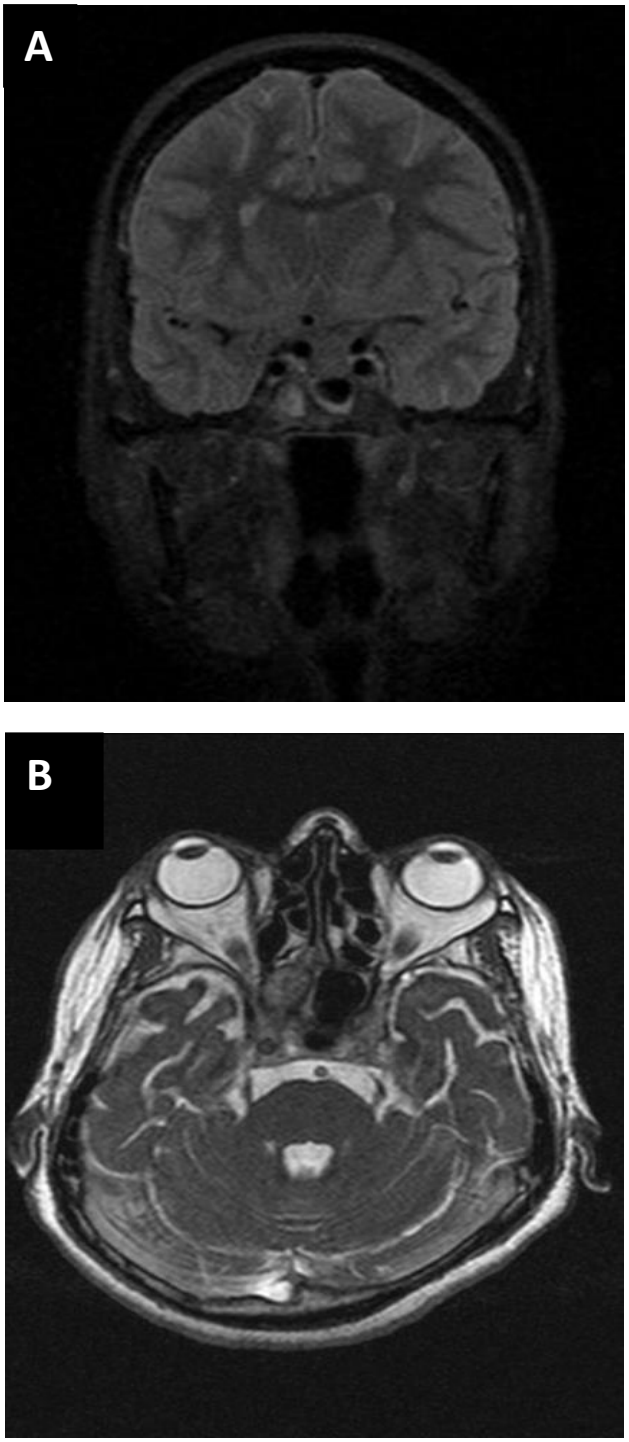

Figure 1 A & B: Magnetic resonance imaging showing mucosal disease in right sphenoid with lateral bulge into the right cavernous sinus.

Right sphenoidotomy was done and samples were taken for histopathologic examination and microbiologic culture. Histopathology showed fungal hyphae and spores in the submucosa. Microbiological examination of the specimen revealed broad branching septate hyphae (Figure 2 A) Fungal hyphae and spores in silver staining and (Figure 2 B) scopulariopsis in high power field is seen.

The organism was identified as a Scopulariopsis species on culture, which was sensitive to itraconazole. The patient received 700 mg (50 mg daily) of intravenous Amphotericin B over the next 14 days and oral itraconazole 200 mg per day for 3 months. Follow-up examination were performed twice monthly for the next 3 months. There was complete recovery of ptosis and ophthalmoplegia; however, her vision remained the same.

DISCUSSION

Most cases of invasive infection with Scopulariopsis have been seen in patients with hematologic malignancies or in recipients of allogeneic bone marrow transplant. Neglia et al.[5] described two immunosuppressed patients who had Scopulariopsis infection of the nasal septum and mastoid tissue that did not respond to aggressive surgical debridement and systemic antifungal therapy. At autopsy, both patients had residual disease, one with widespread involvement of the brain and lungs. Similarly, Scopulariopsis infection of lungs,[5] the sinonasal passages, with dissemination,[6] and the great toe, causing osteomyelitis[1] have documented in immunosuppressed hosts. Immunosuppressed patients have generally responded poorly to antifungal therapy. However, a recent report of invasive sinonasal disease caused by Scopulariopsis in a child with Hodgkin's disease, a combination of surgery and therapy with amphotericin B and itraconazole eradicated the infection completely.[7]

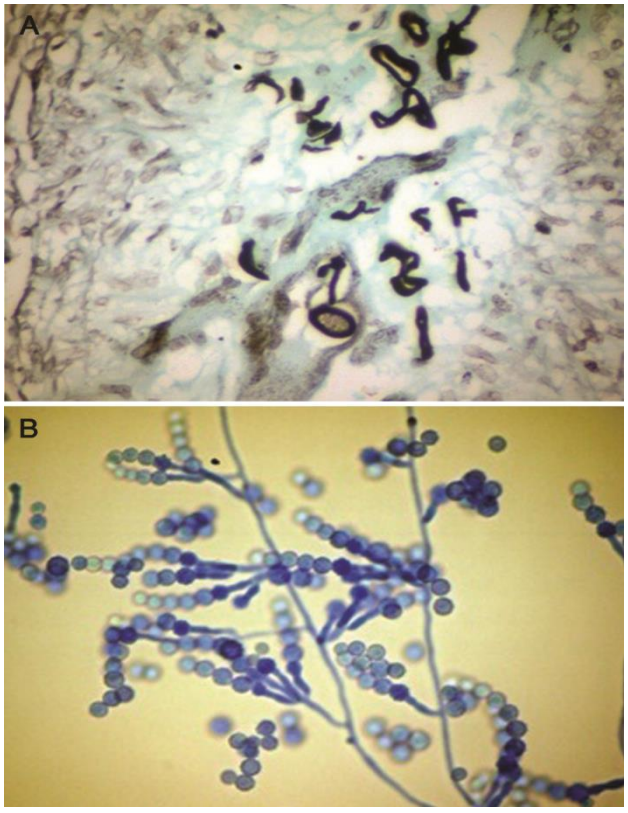

Figure 2: Microbiological examination of the specimen A) fungal hyphae and spores in silver staining, B) scopulariopsis in high power field.

The present report describes a case of orbital infection caused by scopulariopsis in a diabetic patient. In this case report, the patient's presentation was mimicked with that of orbital mucormycosis, though the patient was not in diabetic ketoacidosis.

Infection with Scopulariopsis in immune-compromised patients has resulted in disseminated infection and death or locally persistent infection at the time of autopsy despite aggressive therapy with Amphotericin B. To our knowledge, scopulariopsis has not previously been identified in the literature as a cause of orbital infection, although prior reports have associated Scopulariopsis with endophthalmitis and keratitis.[8]

The optimal antimicrobial regimen for treating scopulariopsis infection is unknown. Results from in vitro susceptibility test vary widely, but the organism is often highly resistant to usual array of agents used to treat systemic fungal infections. Two isolates of scopulariopsis that caused fatal disseminated disease were resistant or only moderately susceptible to Amphotericin B.[5] Other studies have also suggested that the organism may be resistant to amphotericin B.[8-10] In contrast with previously described patients with Scopulariopsis species infection, our patient responded very well to intravenous amphotericin B, suggesting that in vitro susceptibility testing of imidazole might not correlate with clinical outcome.

The case described herein demonstrates the expanding spectrum of fungal organism that may cause orbital infection in diabetic patients. Presentations of scopulariopsis usually mimic that of mucormycosis. Hence, early diagnosis using culture and in-vitro susceptibility testing of the isolates to itraconazole and miconazole is inevitable for the complete eradication of the infection.

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