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CASE REPORT

Sino-orbital Aspergillosis: A Case Report and Brief Review of Literature

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ABSTRACT

Aspergillosis of head and neck region primarily affects the nose and paranasal sinuses. Any type of paranasal aspergillosis may progress to more aggressive disease illustrating the importance of early recognition of this increasingly encountered disease. The invasive and in particular, the fulminant forms are associated with high mortality. We report a case of invasive aspergillosis of right maxillary sinus and orbit in an immunocompetent individual, along with a critical review and update of the literature. The patient underwent surgery for the debridement of right maxillary sinus through a Caldwell-Luc approach and drainage of orbital abscess as well followed by intravenous therapy of amphotericin B. Recovery with reduction of all the signs and symptoms was seen after 38 days from the appearance of first symptoms. Successful treatment of aspergillosis requires prompt diagnosis and rapid institution of therapy, because delay or nonaggressive therapy can result in the spread of infection with lethal consequences.

Keywords: Fungal infection, Paranasal sinuses, Invasive aspergillosis.

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INTRODUCTON

Aspergillosis belongs to the category of systemic mycoses. Though rare in immunocompetent individuals, there is an increased incidence of invasive aspergillosis in the immunocompromised patients, and its incidence has only increased over the past two decades.[1-3] Although pulmonary invasive aspergillosis is most common, other anatomical sites have been described as rare including the skin, sinuses, cerebral meninges, myocardium, liver, thyroid, renal tissues and bone.[4,5] Primary invasive aspergillosis is also rare, and there is limited information addressing this problem.

The invasive and in particular, the fulminant forms are associated with high mortality. Invasive form primarily affects nose and paranasal sinuses. Orbital involvement worsens the prognosis because of ready availability of pathways for further intracranial spread, such as superior orbital fissure, optic canal that directly open into the middle cranial fossa. A high degree of suspicion for the diagnosis and early aggressive therapy are suggested to decrease morbidity and mortality. Continued data collection and research are needed for different diagnostic and treatment modalities for invasive aspergillosis. We report a case of invasive aspergillosis in an immunocompetent individual, along with a critical review and update of the literature regarding the differential diagnosis, histopathology and treatment options.

CASE REPORT

A 60-year-old healthy male presented with a 2 weeks history of throbbing, intermittent toothache. He had later developed pain and swelling in the infraorbital region on the right side of the face radiating to the temporal region on that side since 3 days. On examination, there was grossly decayed tooth 16, swelling and tenderness in right infraorbital region. On the basis of clinical presentation, a diagnosis of dentoalveolar abscess with infraorbital cellulitis was made. Tooth 16 was extracted and pus was drained. Two days later the patient came back with increased pain and swelling. He had periorbital swelling, chemosis and lacrimation (Figs 1 and 2). The visual acuity and fields were normal. Differential diagnosis of cavernous sinus thrombosis, acute sinusitis with orbital cellulitis and orbital abscess was made and empirical antibiotic therapy was started. X-ray Waters' view and computed tomographic (CT) scan were advised. Waters view revealed haziness of the right maxillary sinus (Fig. 3) and CT scan revealed right maxillary sinus opacity. The opacity is seen to extend into the ipsilateral nasal cavity and evidence of hypodense lesion with few air pockets in the floor of the orbit extending into subcutaneous plane of the periorbital region on right side and proptosis of the right eye (Figs 4 and 5).

The patient subsequently underwent second surgery for the debridement of right maxillary sinus trough a Caldwell-Luc approach and drainage of orbital abscess as well. The grayish-yellow pus with some necrotic material obtained following surgery was sent for microscopy with potassium hydroxide (KOH) and fungal culture. The culture demonstrated branching hyphae of *Aspergillus fumigatus*. Intravenous therapy of amphotericin-B was administered with 50 mg of amphotericin-B over a period of 10 days. Recovery with reduction of all the signs and symptoms was seen after 38 days from the appearance of first symptoms (Figs 6 and 7).

Fig. 1: Preoperative clinical presentation

Fig. 2: Preoperative clinical presentation profile view

Fig. 3: Preoperative Water's view

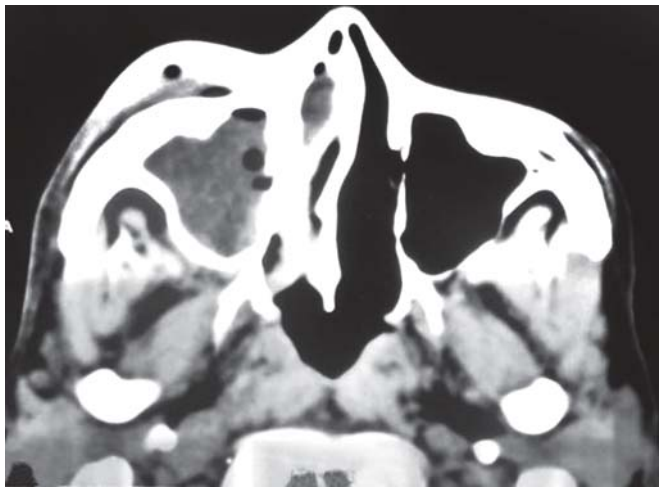

Fig. 4: Preoperative CT scan showing hypodense area in the right maxillary sinus and nasal cavity

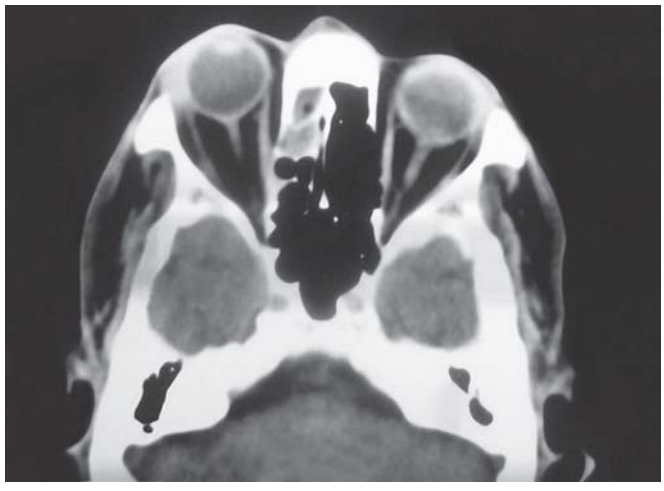

Fig. 5: Preoperative CT scan showing right orbital proptosis

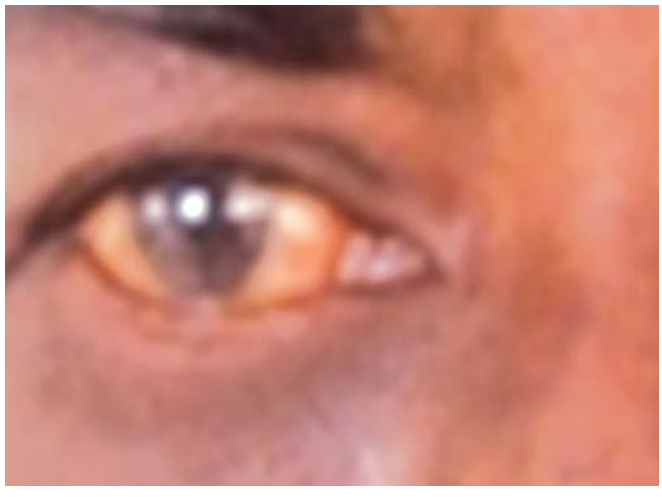

Fig. 6: Postoperative clinical picture showing recovered abscess

DISCUSSION

Aspergillus species are ubiquitous, commonly found in the soil and decaying vegetation. The spores are typically inhaled or ingested without consequence in the normal host and exposure to this fungus is frequent, yet disease due to tissue invasion is uncommon in the immunocompetent host.[1] The portals of entry for *Aspergillus* include the respiratory tract, damaged skin or other operative wounds, the cornea and the ear.[6] An intact immune system can prevent the disease in a healthy individual and there may be mechanism

Fig. 7: Postoperative X-ray showing clear right sinus

by which fungi effect on the sinus mucosa in susceptible individuals only.[7]

Aspergillosis is the second most frequently seen fungal infection of the face and mouth in patients receiving chemotherapy. It is second to *Candida* in its frequency. The overall case fatality rate of 58 to 67% demonstrates that invasive aspergillosis of any type remains a highly lethal opportunistic infection despite the availability of newer antifungal therapies and improved management of underlying diseases.[1,6,8]

Aspergillosis of head and neck region, primarily affects the nasal cavity and paranasal sinuses. *Aspergillus* as a pathogen cannot actively penetrate undamaged and intact mucus membrane or skin as it lacks keratolytic enzymes. On the basis of this finding, paranasal sinus aspergillosis is now classified into invasive (acute fulminant, chronic invasive, granulomatous invasive) and noninvasive (fungus ball and allergic fungal rhinosinusitis) forms with their own pathophysiology and clinical presentation.[8] Any type of paranasal aspergillosis may progress to more aggressive disease illustrating the importance of early recognition of this increasingly encountered disease. [9,10]

Invasive aspergillosis of the sinuses with involvement of adjacent structures is a well-documented cause of morbidity and mortality in immunocompromised patients with the primary risk factors being neutrophil defects and corticosteroid use.[1] Other predisposing factors include HIV infection, diabetes mellitus, use of prosthetic devices or trauma, excessive environmental exposure and possibly advanced age.[2,11,12] Orbital involvement in such patients is known to cause loss of vision. Orbital involvement occurs by contiguous spread of the disease from paranasal sinuses, by expansion or bone erosion due to pressure effect of the polyps or fungal tissue invasion. It is considered to worsen the prognosis of sinonasal aspergillosis. Involvement of the CNS is present in 10 to 15% of patients with disseminated diseases and has mortality rate of 40% within these patients.[13-15]

Rarely has invasive *Aspergillus* infection been described in immunocompetent patients. In a literature review of the last 30 years, we found only 16 cases of sino-orbital aspergillosis (not involving central nervous system) in healthy individuals.[2,15] The extension of the sinus infection is either direct or through osseous structures like the lamina papyracea or through hematogenous spread by valveless venous plexus to the orbit, brain or skin.[16] Localized, invasive sino-orbital aspergillosis is a progressive, relentless condition and has a fatal outcome. Early diagnosis of orbital involvement in patients with fungal sinusitis remains a challenge. The presentation of sino-orbital aspergillosis can mimic infectious diseases like mucormycosis, neoplastic, vascular and neuro-ophthalmic diseases.

Our patient was immunocompetent and presented with painful decayed tooth and infraorbital swelling, misleading us toward dentoalveolar abscess with infraorbital cellulitis. However, the rapid progression of his condition with orbital involvement and severe unilateral headache raised a suspicion of sino-orbital aspergillosis. Fortunately, he had neither ophthalmoplegia nor visual loss, which is commonly noticed in invasive sino-orbital aspergillosis. The CT findings and the presence of hyphae in the KOH preparations of the drained material potentiated our suspicion and was confirmed by fungal culture. Unlike most patients with sino-orbital aspergillosis who have mainly sphenoidal sinus involvement, our patient had maxillary sinus aspergillosis with orbital invasion.

There are very few well-conducted, controlled clinical trials for the treatment of invasive aspergillosis, and none specifically aimed at the treatment of primary invasive aspergillosis. It has been suggested that the treatment of invasive aspergillosis is surgical and medical. Antifungal therapy or surgical therapy alone can produce suboptimal results. Successful treatment of aspergillosis requires prompt diagnosis and rapid institution of therapy, because delay or nonaggressive therapy can result in the spread of infection with lethal consequences.

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