

Orbital Invasive Aspergillosis - A Rare Fungal Infection of Eye

Saksham Nahar¹, Ajay Kumar Pillai², J. Arun Kumar³, Neha Goyal⁴, Santosh Kumar Singh⁵

1 Post Graduate Student, Department of OMFS, Peoples Dental Academy, Bhopal (M.P.), India. 2 MDS, Associate Professor, Department of OMFS, Peoples Dental Academy, Bhopal (M.P.), India. 3 MDS, Associate Professor, Department of OMFS, VMSDC, Salem (T.N.), India. 4 Clinician, Chaudhary Hospital, Bhopal (M.P.), India. 5 MDS, Senior Lecturer, Department of Endodontics, Peoples Dental Academy, Bhopal (M.P.), India.

Abstract

Invasive orbital aspergillosis in immunocompetent patients is a rare clinical entity and is often misdiagnosed. Infections of the eye give rise to severe ocular morbidity. Morbidity and Mortality may be avoided with timely treatment. We present the clinical features, treatment and outcome of a 34 year old male patient who is diagnosed for having fungal infection aspergillosis of the left apex of the orbit. A radiopacification of the left orbital apex was observed in CT scan for which biopsy was performed and diagnosis was made as Aspergillosis.

Key Words: Fungal Infection, Aspergillosis, Fungal Eye Infection.

Introduction:

Invasive Aspergillosis in a healthy host is a rare occurrence but it may become pathogenic under special circumstances in head and neck region. Aspergillosis of paranasal sinus is one of the infrequently reported diseases which can occur as invasive and non – invasive forms. Diagnosis of the condition itself is a difficult task and challenge for the clinician. Early diagnosis is essential in order to avoid high morbidity and mortality associated with the destructive disease and to instigate treatment before irreversible condition arise. The purpose of this paper is to report a case of fungal infections involving the orbital region in immune-competent patients with an emphasis on the fact that early diagnosis is vital in these infections, because delay in initiation of treatment can be life threatening due to propensity of fungi to invade adjacent blood vessels and embolize to distant organs.

Aspergillus is pathogenic in birds, animal and man. Though Aspergillus Fumigatus is the most common fungi which affect the paranasal sinus, it is rarely reported in the English literature. Mycotic sinusitis is on the increase worldwide. It is unclear whether this increase is due to a heightened awareness of the disease or rather to a higher incidence

precipitated by the increased use of antibiotics and corticosteroids.¹

This fungus grows best anaerobically. The Primary sites of fungal infection are respiratory tract, external auditory canal, naso-pharynx, cornea, gastrointestinal tract and occasionally the skin. Opportunistic fungal infections usually occur in immune-compromised patients, but can infect healthy individuals as well.²⁻⁵

This paper reports a case of aspergillosis in an apparently healthy male with involvement of the apex of the orbit.

Case Presentation:

A 34 year old male patient presented with chronic headache and proptosis of left eye (Fig 1). The visual acuity and fields were normal. His medical history, inspection and palpation were unremarkable and the patient was found to be immuno-competent.

As a radiographic investigation computed tomography (CT) scan of orbit was taken, which revealed an ill-defined, heterogeneous, hyperdense mass infiltrating the orbital apex (Fig 2&3), Idiopathic orbital inflammatory syndrome, optic neuritis and orbital bacterial cellulitis/orbital abscess

cellulitis were counted as the differential diagnosis for the same.

Patient was undertaken for biopsy and the histopathological examination of specimen showed tissue invasion by fungal hyphae, with characteristics

of *Aspergillus*: non-caseating multinucleate giant cell granulomas, and chronic inflammatory infiltrate composed of eosinophils, lymphocytes and plasma cells. Culture on Sabouraud's agar media was positive grew *Aspergillus flavus* (Fig 4&5).



Figure No. 1: Photography showing proptosis of left eye



Figure No. 2: Computer tomography (CT) scan of the orbit showing heterogeneous mass in the apex of the orbit, of the right maxillary sinus with evidence of palatal perforation.



Figure No. 3: Computer tomography (CT) scan of the orbit showing heterogeneous mass in the apex of the orbit.



Figure No. 4: Exposure of the lesion through an orbital incision



Figure No. 5: Specimen of excised necrotic maxilla

Treatment:

The patient was hospitalized and extensive debridement of the orbital apex under general anaesthesia was performed. Surgical exploration was performed by infra orbital incision (Fig 6). The orbit was explored and friable granulomatous material was found to extend to the apex of the orbit. Extensive debridement was done (Fig 7) and primary

reconstruction was done. The patient was administered Amphotericin-B, 0.8mg/kg/day, intravenously for two weeks. Blood urea and creatinine levels were monitored as the drug can cause renal toxicity.

Post operatively the healing was uneventful (Fig 8) and follow up examination of the patient's clinical outcome was found to be satisfactory.

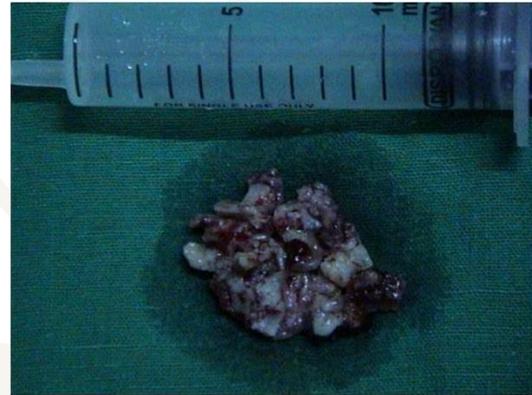
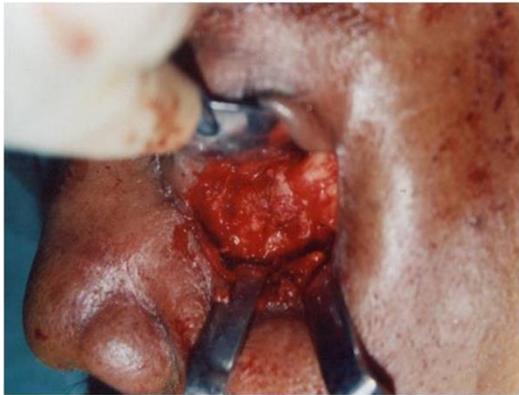


Figure No. 6 & 7: Hematoxylin and eosin stained section showing fungal hyphae with spores.



Figure No. 8: Post operative follow up after 8 months

Discussion:

Aspergillosis is a large spectrum of diseases caused by members of the genus *Aspergillus*. It is a saprophytic mold of the Ascomycetes class that is widely distributed in the environment and is particularly found in soil, cereals, and decaying

vegetation. Although invasive fungal sinusitis may be seen in apparently healthy individuals,¹⁻³ *Aspergillus fumigatus* is most often encountered in among immune-compromised patients.³

Aspergillosis is the most common fungal infection of the para-nasal sinuses in both healthy and

immune compromised subjects. Two ways of contaminating the para-nasal sinuses with aspergillus exists. The first is the “aerogenic” pathway in which spores are inhaled directly in the antrum and the second is “iatrogenic” mode where spores are introduced to the antrum via an oro-antral communication that occurs at the time of a dental procedure such as a dental extraction or a root canal perforation. Once the spores are introduced, they may act as opportunistic pathogens and colonize in the maxillary sinus, particularly when conditions that decrease sinus ventilation, such as bacterial sinusitis already exist. The individual species *A. fumigatus*, *A. flavus*, *A. glaucis*, *A. terreus*, and *A. Niger* have been implicated in human infections, with *A. fumigatus* being the most common causative agent.⁴

Aspergillosis appears in three forms: 1) saprophytic, in which there is fungal growth without invasion of viable tissue; 2) allergic, characterized by the presence of a hypersensitivity reaction to fungal hyphae or conidia; and 3) invasive, in which there is extension of the fungus into viable tissue, resulting in severe necrosis.⁵ The saprophytic and allergic forms affect the immune-competent host and have relatively low morbidity and mortality.⁶ where as invasive or fulminant aspergillosis is rapidly progressive and primarily affects patients who are immune-compromised, such as those with diabetes mellitus or a malignant disease or those undergoing steroid and immunosuppressive therapy.^{7, 8}

Allergic aspergillosis sinusitis was first described by Katzenstein et al in 1984.⁶ A very rare case of aspergillus infection of the periodontal tissues and dental pulp, confirmed histologically, was reported in a tuberculosis patient by Kobayakana et al (1964). Upon review of the English-language literature, Thomas H. Faqua et al found only 25 reported cases of primary intra oral aspergillosis.¹¹

Usually only one sinus, i.e. Maxillary sinus, is affected, and bacterial super infection may lead to episode of acute sinusitis. Symptoms such as pain and swelling, ulceration, nasal obstruction and rhinorrhea may also develop with the development of ocular and neurologic signs occurring from either local compression or direct invasion, hence the clinical

picture of para-nasal aspergillosis can therefore be similar to the malignant disease, chronic sinusitis with osteomyelitis, Wegener’s granulomatosis, mucormycosis, and inverted.

Because the clinical picture of aspergillosis is often similar to that of other disease, effective imaging techniques are essential to help establish an early diagnosis. The presence of a radiodense foci in associated with homogenous opacification of the sinus is highly suggestive of a non-invasive mycetoma. In addition it is found to affect only one sinus at a time. Such foci may simulate either a foreign body, an antrolith, a calcified hematoma, or an osteoma.¹⁰

On a radiographic evaluation, invasive form usually involves multiple sinuses with possible added evidence of bony destruction. Allergic aspergillosis presents with diffuse expansile involvement of multiple sinuses. Tissue invasion is not a common characteristics however, cases involving intracranial extension have been reported.¹¹

CT examination is a more sensitive diagnostic tool than plain radiography, and in addition to being useful in identifying early invasive disease.

Oral lesions of aspergillosis, seen predominantly in some immune-compromised patients, are yellow or black, necrotic ulcers, typically in the palate or occasionally the posterior tongue. The main differential diagnosis are from mucormycosis, pseudomonas.

Conclusion:

Fever and incorrect initial diagnosis was found to be associated with high mortality rate in patients with invasive aspergillum of the orbit. The most common ocular symptoms were visual disturbance, periorbital swelling and periorbital pain. Clinical, radiological and histopathological findings are useful for establishing the extent of the disease and eliminating other conditions from the differential diagnosis.

Key message:

1. Fungal infections of the paranasal sinuses and orbit are usually seen in immunocompromised and occasionally in immune-

competent individuals, so clinicians should be aware that chronic sinusitis that is unresponsive to usual management is highly suggestive of mycoses of the paranasal sinuses.

2. The bone of maxilla-facial region rarely undergoes necrosis due to its rich vascularity. Unlike bacterial infections which cause tissue necrosis due to its direct effects, the destruction caused by fungi is due to avascular necrosis secondary to embolisation of the blood vessels.

References:

1. Taneja T., Saxena S., Pandey A., Bansal V., Aggarawal P. Aspergillosis involving the maxillary sinus-a report of two cases and a brief literature review. *Int J Oral-Med Sci*, 2010; 9(1): 42-47.
2. Washburn RG, Kennedy DW, Begley MG, Bennett JE: Chronic fungal sinusitis in apparently normal hosts. *Medicine(Baltimore)*, 1988; 67: 231-247,.
3. Milory CM, Blanshard J, Slucas, Michaels L: *J clin pathol*. 1989; 42: 123-127
4. Young RC, Bennet JE, Vogel CL: Aspergillosis: The spectrum of the disease in 98 patients *Medicine(Baltimore)* 1970; 49: 147-173,.
5. Fraser RS: Pulmonary aspergillosis: Pathologic and pathogenic features. *PatholAnnu*. 1993; 28: 231-277.
6. Katzenstein ALA, Sale SR, Greenberger PA: Pathologic findings in allergic sinusitis: A newly recognized form of sinusitis. *Am J SurgPathol*, 1983; 7: 439-443.
7. Myoken Y, Sugata T, Tai Chi K, Fujihara M: Pathologic features of invasive oral. *J Oral Maxillofac Surg*. 1996; 56: 263-270.
8. Dreizen S, Bodey GP, McCredie KB, Keating MJ: Orofacial aspergillosis in acute leukemia. *Oral Surg, Oral Med, Oral Pathol*. 1985; 59: 499-504.
9. Katzenstein A, Sale S, Greenberger P: Allergic aspergillosis sinusitis: a newly recognized form of sinusitis. *J Allergy Clin Immunol*. 1983; 72: 89-93.
10. Garcia MF, Pinilla JC, Echeverria CL, Ferrero AE, Hernandez AV: Invasive maxillary aspergillosis: Report of a case and review of the literature. *Medicina Oral*. 2000; 7: 200-205.
11. Fuqua TH Jr, Sittitavornwong S, Knoll M, Said-Al-Naief N: primary invasive oral Aspergillosis: An updated literature review. *J oral maxillofac Surg*. 2010; 68: 2557-2563.

Corresponding Author

Dr. Saksham Nahar

Department of OMFS, Peoples Dental Academy, Bhopal (M.P.), India.

Email id- saksham3dec@gmail.com