

# ASPERGILLOSIS OF THE MAXIL-LARY ANTRUM: A CASE REPORT

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## ABSTRACT

Aspergillosis (a fungal infection) of the nasal and paranasal sinuses is recognised as being second to candidiasis, among opportunistic infections in immunocompromised patients. It ranges from allergic sinusitis, fungus balls to fulminating infections. However invasive infection in normal hosts is a very rare occurrence. We report one such case which had caused extensive destruction and clinically simulated malignancy.

**Key words:** aspergillosis, fungal, invasive, fulminant, granuloma

## INTRODUCTION

Aspergillosis predominantly an opportunistic fungal disease is caused either by sensitisation to or parasitic colonization of or tissue invasion by species of genus *Aspergillus* with *A. fumigatus*, *A. flavus* and to some extent *A. niger* being pathogenic (Martinez 1992)<sup>1</sup>.

Paranasal and nasal Aspergillosis has been recognized in many forms and was initially broadly classified as non invasive and invasive forms (Hora 1965)<sup>2</sup>.

But Sarti and Lucenten<sup>3</sup> in 1988 classified Aspergillosis as:

- Allergic *Aspergillus* sinusitis
- Non invasive type (fungus balls)
- Invasive type (akin to a malignant neoplasm with proptosis and facial mass)
- Fulminant type (which are angio invasive, rapidly destructive and lethal).

The recent classification has been put forth in 1994 by Rowe-Jones<sup>4</sup>. They have classified Aspergillosis into 3 main types: Non-invasive, invasive and destructive non-invasive types, which is further

classified into Aspergilloma / fungus ball / Mycetoma (usually affecting one sinus) or allergic *Aspergillus* sinusitis (involving more than one sinus). Invasive type represents true fungal tissue invasion that can be either slowly progressive and destructive (non fulminant) or highly aggressive and lethal fulminant. Destructive non-invasive type (semi-invasive) is locally destructive but shows no tissue invasion.

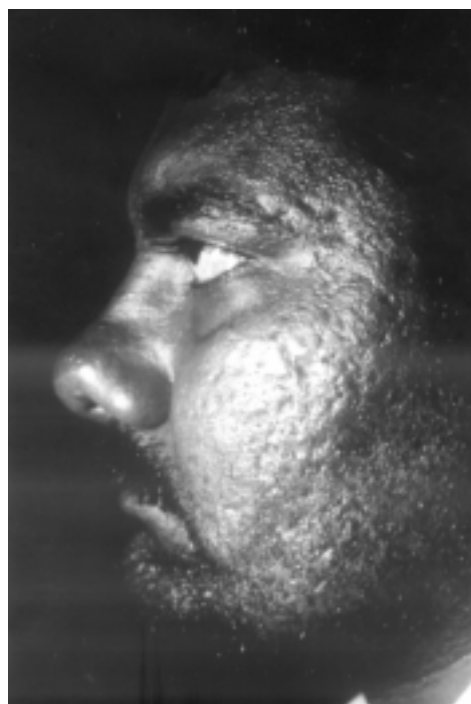


Fig 1: Photograph of the patient showing diffuse swelling in the left maxillary region with proptosis.

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Fig. 2: CT scan image showing hyperdense mass in the left infraorbital region with breach in the infraorbital margin.

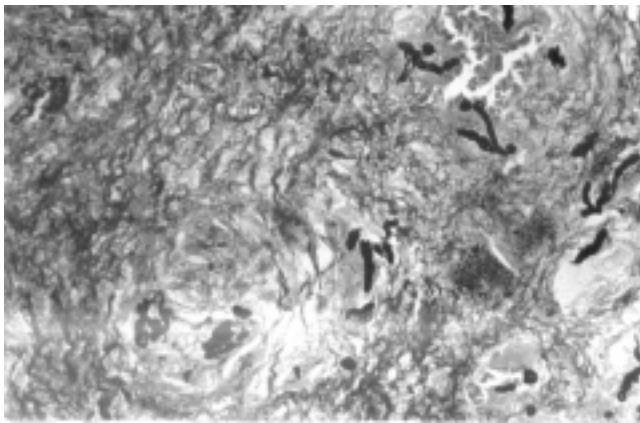


Fig. 3: Photomicrograph showing scattered multinucleated giant cells, some Langhan's type (arrows) in granuloma formations with dense lymphocytic infiltration (H&E x25)

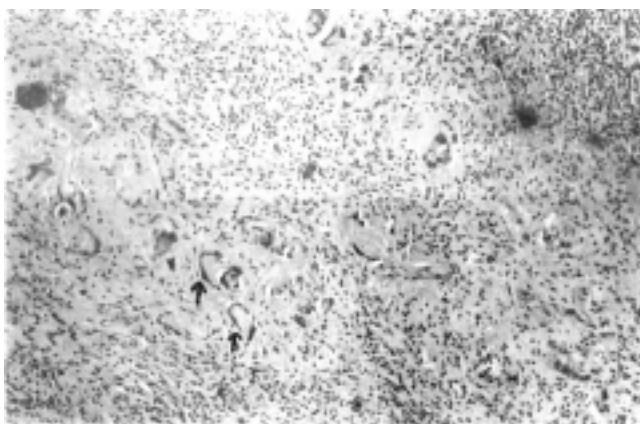


Fig. 4: Photomicrograph of the aspergillum organisms in the granulomas showing typical branching septate hyphae (Methenamine Silver x45)

Aspergillus sinusitis in normal hosts is a common occurrence but invasive and fulminant types are common in immunocompromised patients. However invasive forms in normal hosts is very rare. Here we report a case of invasive aspergillosis that occurred in a healthy host.

### CASE REPORT

A 21 yr old male patient was seen at our out patient department in December 1997, with a complaint of a slow growing swelling in the lower left eyelid of 8 months duration. Past history revealed that he had been operated in 1994 for a right antral polyp. Routine blood examination was within normal limits and HIV status was negative. Radiographic examination showed opacification of the left maxillary sinus. On examination a firm swelling measuring approximately about 2 x 1.5 cm was observed in the infraorbital region.

Further, the patient underwent total excision of the mass through a blepharoplasty incision of the left maxillary sinus. A diagnosis of non specific granuloma was given. The sinus involvement was thought to be bacterial sinusitis. Again in August 2000 the patient reported with disturbed left eye vision accompanied by mild infraorbital pain and a squint. On clinical examination a diffuse swelling over the left maxillary region, and an evident proptosis of the same side was observed (Fig. 1).

CT scan showed a hyperdense expansile mass occupying the left maxillary antrum causing destruction of the infraorbital wall (Fig. 2) and extending below and behind the left orbit, with proptosis of the eyeball. It was seen involving the whole of left ethmoidal cell complex and also partially extending to sphenoidal sinus. However the involvement on the right side was minimal and showed only mucosal thickening.

On surgical exploration of the sinus, there was considerable amount of greenish black necrotic debris filling the sinus. Pathological examination revealed Aspergillosis of the paranasal sinus. The patient was put on oral antifungal and discharged,

as further surgery was not considered to be appropriate.

## HISTOPATHOLOGICAL EXAMINATION

Examination of haematoxylin and eosin stained sections revealed a fibro-cellular connective tissue with several non-caseating granulomas and many Langhan's type and foreign body giant cells (Fig. 3) and many epithelioid cells. An amorphous material was present in the cytoplasm of giant cells suggestive of foreign body with occasional eosinophilic infiltration was observed. Further staining with methenamine silver stain showed septate dichomatous branching hyphae suggestive of *Aspergillus* fungus (Fig. 4). However, AFB stained sections, to rule out TB, were found to be negative.

## DISCUSSION

Aspergillosis, a spore forming fungus, first identified by Sluyter in 1847, is an ubiquitous organism and thrives in soil, water and decaying organic debris (Hinson, 1952)<sup>5</sup>. Schubert in 1985 was the first to describe aspergillosis of nasal and paranasal sinus (Stammberger 1984)<sup>6</sup>. Air born spores are introduced by inhalation or during surgical procedures including even tooth extractions. They are frequent inhabitants of human respiratory tract. The most frequent sites are the lung, sinus and nasal cavity with oral cavity, external ear canal, meninges, spleen and bone also being involved.

*Aspergillus flavus* is considered to cause more destruction in paranasal sinus and oral cavity, compared to *aspergillus fumigatus* because of its potent toxins which enables the former to penetrate the mucosa (Shannon 1990)<sup>7</sup>. Paranasal Aspergillosis is considered as a spectrum of diseases encompassing many forms. It can become localized or become destructive and invasive and extend to intracranial structures or even extend to oral cavity and cause palatal perforation. If left untreated, it may progress to the disseminated form. These infections present with signs and symptoms of allergic sinusitis and rhinitis facial pain, unilateral prop-

osis, epistaxis, frequent head aches, greenish discharge etc, and some non invasive and invasive forms can also go unrecognised (Napoli, 1991)<sup>8</sup>. Our patient, however, presented with infraorbital swelling and mild pain, with no other acute symptoms.

Radiographic changes in Aspergillosis usually remain non-diagnostic and are similar to changes seen in that of bacterial sinusitis. Bone destruction can be seen in the invasive forms. Calcifications are sometimes reported to occur. However this was not seen in our case.

Clinically thus the invasive lesions can mimic malignancy or Wegener's granuloma, rhinoscleroma / midline lethal granuloma and confirmation can only be sought after biopsy, with or without culture of fungus. Grossly, the infected tissue exhibits yellowish, brownish, grey or black in colour and is of cheesy consistency, containing dirty or muddy material (Chang et al, 1992)<sup>9</sup>.

Histopathologically, invasive lesions comprise chronic granulomatous reactions and appear like any of the granulomas of sarcoidosis, midline lethal granuloma or foreign body granuloma. If Langhan's type giant cells are seen as in the present case, pre-existing TB granuloma has to be ruled out, as Aspergillosis can also grow in tuberculous cavities. Though hyphal forms can be seen faintly in haematoxylin and eosin stained sections, they may go unnoticed unless selective special stains like PAS or methenamine silver are employed. They appear as septate hyphae with branching at 45° angles and are about 2-4 µm in diameter. This fungus differentiated from mucormycosis where broader nonseptate hyphae with dichomatous branching at 90° angle is observed. As culture may be negative even after employing Sabouraud agar, demonstration of the hyphae in tissue sections are more reliable and conclusive, but species cannot be confirmed.

The patient remained afebrile with no other symptoms and as the blood picture was normal, a vas-

cular dissemination of Aspergillosis was ruled out. As the lesion had caused extensive infiltration into the adjacent sinuses and into the orbit and exhibited tissue invasion, it was categorised as an invasive but locally destructive form of Aspergillosis. Mucormycosis / Zygomycosis can also present; in the same way and has to be considered in the differential diagnosis especially in cases of proptosis.

Early recognition of these diseases and especially differentiating it from malignant lesions and other granulomas of the sinuses are very important as its presentation is very deceptive and misleading.

Treatment depends on the degree and type of Aspergillus infection. Usually it consists of antifungal drugs with concomitant surgery. Steroids are given in case allergic aspergillosis and surgery is usually not indicated.

## CONCLUSION

Invasive slowly progressive Aspergillosis can remain clinically innocuous. We should consider such invasive and destructive clinical presentation especially with proptosis in a broader sense and help clinicians in instituting proper treatment at appropriate time and help prevent such infection progressing into lethal and fulminant forms.

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