Invasive Maxillary Aspergillosis in an Otherwise Healthy Individual

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Introduction
Aspergillus is a spore-forming fungus found in soil, plants and decaying vegetation. It can involve respiratory tract, paranasal sinuses, ears, liver, spleen, bone, etc. both in healthy and immunocompromised hosts. Aspergillosis of the paranasal sinuses can occur as a noninvasive, invasive, fulminant and allergic infection. Invasive aspergillosis is usually seen in immunocompromised individuals. Gerson, et al. observed that prolonged granulocytopenia (neutropenia) was the only statistically significant risk factor for the development of invasive aspergillosis in the pulmonary tree, while invasiveness is rarely observed in normal hosts.

We are reporting a case of aspergillosis of the maxillary sinus extending to the anterior cranial fossa in an otherwise healthy individual.

Case Report:
A 56-year-old housewife developed right-sided headache and proptosis one month previously. There was no complaint of defective vision, diplopia, nasal obstruction, discharge from nose, sore throat or ear pain. There was no history of diabetes, evidence of HIV risk factors such as blood transfusion, homosexuality or intravenous drug use, etc. On examination, cardiovascular, respiratory, and central nervous systems were normal. Examination of the nose revealed a deviated nasal septum and a mass in the middle meatus. Right-sided proptosis was present, but no nystagmus, pupillary and fundal abnormalities were seen. Visual acuity was normal in both eyes, and full range of movements of both eyes, though painful, was possible. Computerized tomography revealed a mass in the ethmoid and sphenoid sinuses extending into the orbit. No intracranial extension of the tumor was observed. Blood glucose (fasting and post-prandial), urea, creatinine, chest x-ray, routine hemogram, and urinalysis were all within normal limits. HIV by ELISA was negative. Biopsy of the mass showed diffuse collection of giant cells, lymphocytes and numerous septate branching hyphae (fig. 1). Culture of the biopsy material was sterile aerobically and anaerobically for bacteria. However, Aspergillus niger grew on Sabouraud's dextrose agar (fig. 2), which was identified by standard methods. The patient was treated with amphotericin B in the dose of 1.0 mg per kg per day intravenously as soon as the biopsy report and culture report were obtained (before surgery). The patient underwent right medial maxillectomy through lateral rhinotomy approach for better visualization of maxillary, ethmoid and sphenoid sinuses. The tumor extended upwards, eroding the cribiform plate. The medial orbital wall was dehiscent and tumor mass bulged into the orbit. Orbital periosteum was intact. Part of the mass was removed, sent for histopathology and culture of fungus. The histopathology and culture of fungus confirmed the diagnosis. A repeat CT done one week later showed a mass involving the sphenoid sinus and spreading to the frontal area with dural involvement (fig. 3 & fig. 4). One week later a right-sided craniofacial resection was performed. The previous rhinotomy incision was opened and maxillary antral contents sucked out; the sphenoidal sinus was opened with gouge and hammer and the contents curetted out. Dural involvement was cleared by resecting the diseased dura, which was sent for histopathologic examination and culture. Temporalis fascia graft was placed over the dural gap and stitched in place. Gel foam was placed over the graft. Dura was stitched to the periosteum over the calvarium and closed with continuous stitch. The antrum was packed and lateral rhinotomy incision was closed in two layers. The histopathology and culture report for fungus corroborated the presence Aspergillus niger for the third time. The patient was continued on amphotericin B; however, on the tenth post-operative day, the patient died due to cerebral edema.
INVASIVE MAXILLARY ASPERGILLOSIS IN AN OTHERWISE HEALTHY INDIVIDUAL

Discussion
Aspergillosis of the paranasal sinus and nose in a healthy individual is being more frequently recognized. The invasive form of this infection is commonly seen in immunocompromised hosts. In this case, the invasive form of aspergillosis was seen in an otherwise healthy individual.

The spores of aspergillus, abundant in the environment, readily disseminate into the air by wind currents. Man gets infected by inhalation, inoculation and by contact. Probably the mode of entry in our case was by inhalation. There are many species of Aspergillus including A. fumigatus, A. flavus, A. niger, A. oryzae and A. nidulans. A. fumigatus is the most common pathogen while it is A. flavus which has been more commonly isolated from paranasal sinus infection. A. niger is rarely reported to cause paranasal sinus infection. To establish the role of aspergillus as pathogen, direct demonstration in tissue and repeated isolation from the clinical materials is mandatory.

As the patient was not immunosuppressed, it is likely that local conditions had predisposed the fungal infection in the maxillary sinus. Stevens has emphasized that functional obstruction of the sinus ostium may result in aspergillus infection and that the fungus favors hypoxic or anaerobic condition. Tissue invasion favors other pathogens such as mucormycosis. Why the aspergillosis should

Figure 1. Microphotograph of biopsy stained with Gromori's Methanamine silver, showing septate hyphae X200.

Figure 2. Microphotograph of slide culture of Aspergillus niger Lactophenol cotton blue X 450.

Figures 3 and 4. CT scan showing mass in the ethmoid, sphenoidal sinus extending to anterior cranial fossa.
become invasive is not known, but duration of infection may be one of the factors.

Treatment depends on the degree of involvement. Occasionally patients who seem otherwise healthy will have recurrent fungal infection in spite of local surgery and may benefit from antifungal drugs. In cases with local tissue invasion, antifungal drugs in addition to surgery are recommended. The drug of choice is amphotericin B, although it has the disadvantage of requiring IV administration and has significant toxicity. The key to successful surgical treatment is the removal of diseased mucosa, adequate aeration and drainage of the involved sinus. Prognosis in otherwise healthy patients treated with surgery and antifungal agents is generally good.

However, in spite of administration of an antifungal drug and extensive debridement surgery, our patient died, as the intracranial extension is associated with high mortality.

Summary

Invasive aspergillosis is usually seen in immunocompromised individuals. We report the case of a 56-year-old immunocompetent housewife with Aspergillus niger invading the ethmoid and sphenoid sinuses and extending to the anterior cranial fossa. A. niger grew on three occasions (from the biopsy, mass and duramater). In spite of medial maxillectomy and ethmoidectomy followed by craniofacial resection and antifungal drugs (amphotericin B) the patient died on tenth post-operative day.

References