Aspergillosis of the Sphenoid Sinus

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• Three of the four previously described patients with isolated aspergillosis of the sphenoid sinus died, owing to intracranial extension of the infection. The case reports of two patients with sphenoid aspergillosis are presented in which surgical exteriorization and aeration of the involved sinus was curative. (Arch Otolaryngol 104:47-50, 1978)

Aspergillus, a fungus of the Ascomycetes class, is worldwide in distribution and is commonly found on decaying debris and in soil. Although man often comes in contact with a variety of species, only seven have been implicated in human infections. Aspergillus fumigatus accounts for approximately 90% of these infections. Aspergillus is perhaps most frequently encountered as a saprophytic growth within the external auditory canal. Aspergillus spores, however, are common contaminants of the respiratory tract, where the most serious infections are also produced.

Aspergillosis may appear as a localized mass within a thin-walled cavity, as a chronic granulomatous disease that resembles tuberculosis, or as an aggressive, invasive infection that is characterized by necrosis, hemorrhage, and systemic dissemination. The latter is usually seen in individuals with underlying debilitating disease or in patients who are receiving irradiation, corticosteroids, antibiotics, or cytotoxic drugs.

Aspergillosis of the paranasal sinuses is rare, except in the Sudan where several reported series indicate that the disease is endemic. The maxillary sinus or the ethmoid complex is infected most commonly. Beginning with Oppe, who in 1897 reported cerebral aspergillosis owing to extension of a sphenoid Aspergillus sinusitis, there have been only four reports of isolated sphenoid sinus involvement. Three reports included fatal termination that was due to CNS involvement. We wish to report two cases of noninvasive sphenoid sinus aspergillosis: one that was discovered as an incidental finding, the other in which the patient had nasal obstruction, post nasal drainage, and bleeding. The condition of both of the patients responded favorably to surgical exteriorization of the involved sinus.

REPORT OF CASES

Case 1.—A 69-year-old man was admitted to the hospital for evaluation of a red lesion behind his left tympanic membrane that was associated with a pulsating tinnitus. The presumptive diagnosis was of a glomus jugular tumor. The patient gave the additional history of chronic, left nasal obstruction, which was attributed to a long-standing deviation of the nasal septum. While the patient was undergoing roentgenographic evaluation of the suspected glomus tumor, an incidental cloudiness of the left sphenoid sinus was discovered (Fig 1). Polytomography of the area showed evidence of bone destruction in the floor of the left sphenoid (Fig 2). Arteriography, which was performed to evaluate the glomus tumor, revealed a large vascular lesion that filled the jugular foramen and extended into the posterior fossa.

Before the start of radiation therapy for treatment of the glomus tumor, the sphenoid lesion was evaluated by surgical exploration that utilized a transseptal approach. Removal of the anterior wall of the sphenoid sinuses revealed the cavity to be filled with a cheesy, grayish-green material. The mucous membrane that lined the sinus was not remarkably thickened, although a small biopsy specimen was secured. Following surgery, the patient noted a great improvement in his breathing that was attributable to the incidental straightening of the nasal septal curvature.

Histological examination of the tissue that had been removed from the sinus showed a mild infiltration of fibroconnective tissue with mononuclear cells, and superficial invasion of that tissue by branching, septate hyphae, which extended from an adjacent mass of fungal hyphae. The morphologic characteristics of the fungal elements were consistent with Aspergillus sp (Fig 3).

The glomus tumor was treated with radiation therapy. At this writing, the patient
Fig 1.—Incidental clouding of left sphenoid (case 1).

Fig 2.—Bone destruction (arrows) in floor of left sphenoid sinus (case 1).

Fig 3.—Case 1. Numerous dichotomous, branching septate hyphae (arrows) with superficial invasion of underlying tissues (Grocott's methanamine silver, × 2,000).

Fig 4.—Case 2. Partial destruction of floor of left sphenoid (arrows).
had a decrease in tinnitus and no evidence of progression of the glomus tumor. Follow-up roentgenograms, taken three years later, revealed only minimal clouding of the previously involved sinus.

Case 2—A 57-year-old woman had a three-year history of bleeding from the posterior aspect of her nose. She also reported nasal obstruction on the left, which usually occurred while she was in the recumbent position, and occasional pain on the left side of the face.

Results of physical examination were within normal limits, with the exception of a deviation of the nasal septum that blocked the left side of her nose. A stream of sanguineous, mucopurulent material was seen on nasopharyngeal examination, and it appeared to originate from high in the left posterior nasal choana.

Roentgenographic examination showed the paranasal sinuses to be clear, except for an increased density in the left sphenoid sinus. Polytomography of the area revealed bone destruction in the floor of this sinus (Fig 4). The sphenoid sinus was approached transseptally under local anesthesia. The orifice of the sinus was filled with a mass of inspissated, brownish mucus that was removed along with the anterior wall of the sinus. The mucous membrane was curetted and appeared to be slightly thickened. The clinical impression was mucocele. Roentgenographic evidence of bone destruction if often seen in patients with this "invasive" form of aspergillosis, while orbital involvement is not uncommon.

The diagnosis of a fungal sinus infection should be suspected whenever a "sinusitis" does not respond to antibiotic therapy or irrigations; when a greenish-white "membrane" is produced by irrigations; when the disease process is confined to a single sinus; or when there is evidence of bone destruction. The differential diagnosis in these patients includes cancer, which often must be ruled out by biopsy.

Specific identification of the agent is desirable even though the histologic morphology allows presumptive identification. Penicillium sp may have similar morphologic features, but it is an unlikely agent. Aspergillus grows readily on simple, fungal isolation media without cycloheximide. Care in the interpretation of cultures of nonbiopsy specimens is required because during periods of fungal sporulation, contaminated respiratory secretions of unaffected patients may produce positive cultures. Laboratory air may also be a source of specimen contamination. A positive culture that is accompanied by a smear showing hyphal fragments, or a heavy, repeated positive culture together with a consistent clinical picture is sufficient for presumptive diagnosis. Positive results for both culture and histological studies of biopsy specimens yield the most definitive conclusions.

Several authors have suggested that Aspergillus fungus becomes pathologic only when the involved sinus becomes relatively anaerobic. In the Sudan, where invasive paranasal aspergillosis is quite common, cultures of the nasal cavity in 19 healthy persons revealed that 18 persons had Aspergillus present, without evidence of pathological involvement. Predisposing factors that may cause a sinus to become relatively anaerobic include nasal polyps, chronic infection, or mucosal thickening owing to allergy. The relative frequency of the disease in the Sudan has been attributed to the dry, dusty climate, and the frequent sandstorms. Because Sudanese tend to seek medical aid rather late, many come to the physician with advanced disease, and a large mass in the maxillary-ethmoid area, with proptosis being commonly present.

Veress el al believed that two additional factors may also play an important role in paranasal sinus aspergillosis. The fungus, given the proper environment, has an inherent ability to invade tissue and release toxic metabolites. Once the fungus has become established, an immunological process also occurs with additional tissue destruction. This immune aspect of invasive aspergillosis is supported by the presence of a fibrinoid necrosis of collagen in the surrounding soft tissues; pseudotubercles similar to those seen in other conditions of delayed hypersensitivity; infiltration of plasma cells and lymphocytes along blood vessels into the adjacent tissues; and the development of vascular alterations similar to those seen in the splenic arteries of patients with systemic lupus and third-stage lues. This arteritis and fibrinoid degeneration of collagen have also been found in primary aspergillosis of the lung and in small arteries of the brain with cerebral aspergillosis. The role of an immunologic response is also supported by the presence of allergic...
cutaneous reactions and a circulating anti-Aspergillus precipitant in the sera of patients with the disease.

Treatment

Paranasal sinus aspergillosis will respond to conservative surgical procedures that establish aeration of the involved sinuses. The Caldwell-Luc approach with the formation of a nasal antral window is usually sufficient when the maxillary sinus alone is involved. If the maxillary-ethmoid complex is infected, then intranasal ethmoidectomy should also be done.

In the Sudan, where orbital invasion and wide tissue involvement is often present, conservative excision of all visible granuloma, along with aeration of the involved sinuses, is usually curative. In these extensive cases it has been found that residual granuloma, found adjacent to the slera or dura, would regress and heal by fibrosis following both excision of the majority of the lesion and a wide drainage procedure. Most authors agree that systemic or topical antifungal agents, when used as the sole therapy for paranasal sinus aspergillosis, are not effective. These drugs have also been used in conjunction with surgical procedures, but their actual contribution to the resolution of the disease has not been established in these cases.

Radiation therapy and desensitization with an extract from the aspergillosis fungus have been also tried without benefit.

Although the conditions of most patients respond to conservative surgical therapy, there have been occasional case reports in which paranasal sinus aspergillosis was a relentlessly progressive disease, despite aggressive therapy. Hora reported a case that was first diagnosed in 1955 as paranasal sinus aspergillosis, which was caused by A flavus. During the intervening six years, the patient required multiple surgical procedures, which were combined with repeated courses of intravenously given amphotericin B. One operative procedure included excision of the granuloma from the apex of the orbit, maxillary sinus, ethmoid sinus, temporal fossa, and pterygomaxillary space. Finally, after six years of intermittent therapy, the patient became symptom-free, and continued to do well for the following four years.

Sphenoid Infections

Isolated sphenoid involvement with the Aspergillus fungus is rare. Four cases have been reported in the literature, three with fatal termination that was due to CNS involvement.

Sood et al described a patient with severe headache, nasal drainage, and decreased visual acuity in her left eye. A transethmoid exteriorization of the sphenoid revealed a chronic Aspergillus infection. The patient died on the fifth postoperative day of CNS complications. A patient who was described by Oppe also died of CNS extension of the infection.

McKee reported the autopsy findings of a 60-year-old man who died from probable cerebral aspergillosis with rupture of the internal carotid artery. McKee believed that the primary site of the fungal infection was the sphenoid sinus.

Kawamoto et al described a 39-year-old woman with a left sphenoid aspergillus infection whose symptom was a bloody postnasal discharge of three months' duration. The patient became symptom-free three weeks after a transnasal surgical procedure to exteriorize the sinus.

Our two patients had deviation of the nasal septum that blocked the left side of their nose. Notably, in both instances only the left sphenoid was involved, which led us to believe that underaeration in the sphenoid recess produced the warm, moist condition that is favorable to growth of the fungus. As in the majority of patients with Aspergillus infection of the maxillary or ethmoid sinuses, a conservative surgical procedure designed to debride and aerate the sinus was curative.

Nonproprietary Name and Trademark of Drug

Amphotericin B—Fungizone.

References