

sicca complex include recurrent otitis media, chronic sinusitis, gingivitis, and dental caries. Systemic complications may be manifested as organ-specific autoimmune disorders.¹

There are no universally accepted criteria for the diagnosis of SS. The diagnosis is established by performing an ophthalmologic examination and a minor salivary gland biopsy. The ophthalmologic examination includes a Schirmer's test and a rose bengal dye test to confirm keratoconjunctivitis sicca. If autoantibodies (i.e., antibody to nuclear antigens, rheumatoid factors, anti-Ro, anti-La) are present, they add confirmation to the diagnosis of SS or related autoimmune diseases. The absence of autoantibodies, however, does not rule out the diagnosis of SS.

Roentgenographic diagnosis of lung complications in SS is greatly aided by CT. Plain chest CT images did not demonstrate lymphomatous changes or bronchiectasis in our patient. High-resolution CT images easily demonstrated bronchiectasis. High-resolution CT has been shown to be a sensitive tool for diagnosing bronchiectasis.⁴

Symptomatic relief and prevention of complica-

tions are important issues in treating SS. Medications or factors that exacerbate the drying of mucous membranes should be discontinued. Dry mucous membranes should be rehydrated. Infections are treated aggressively to prevent complications such as bronchiectasis. Patients with SS require close follow-up because of their propensity to develop lymphoproliferative disorders.^{1, 2, 5} High-resolution chest CT should be performed in patients who have pulmonary complications related to SS.

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***Aspergillus* brain abscess complicating allergic *Aspergillus* sinusitis**

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The ubiquitous fungus *Aspergillus* causes a wide spectrum of clinical syndromes, including allergic bronchopulmonary aspergillosis (ABPA)¹ and allergic *Aspergillus* sinusitis (AAS).² These latter syndromes reflect hypersensitivity responses to fungal antigens in the absence of fungal invasion. The majority of affected individuals are young, atopic, and immunocompetent. We describe a unique case of allergic *Aspergillus* paranasal pansi-

Abbreviations used

- AAS: *Aspergillus* sinusitis
ABPA: Allergic bronchopulmonary aspergillosis
CT: Computed tomography
FEV₁: Forced expiratory volume in 1 second
FVC: Forced vital capacity

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nitis in a patient who had an *Aspergillus* brain abscess and concomitant ABPA.

CASE REPORT

A 32-year-old Indian man with a history of asthma and chronic nasal obstruction who had undergone bilateral Caldwell-Luc sinus drainage procedures in 1986, came to the emergency department in March 1990 after



FIG. 1. Microscopic appearance of the frontal lobe brain lesion. **A**, Fungal hyphae with dichotomous branching characteristic of *Aspergillus* species, eosinophils, and necrotic material. **B**, Charcot-Leyden crystal and necrotic material. (Papanicolaou's stain; original magnification $\times 100$.)

he had a grand mal seizure. Progressive right-eye bulging and frontal headache had been noted for 1 week. He had lived in India until 6 months before admission and had received all of his medical care there. Despite worsening nasal congestion over the 6 months before admission, he had not sought medical attention in the United States. His medications included nasal beclomethasone dipropionate, nasal cromolyn, and a β_2 -adrenergic agonist inhaler. Physical examination revealed the patient to be unarousable with blood pressure of 158/79 mm Hg; pulse, 116 beats/min; respirations, 20/min; and rectal temperature of 38.9° C. He had bilateral nasal polyps and moderate right-eye proptosis without periorbital edema. Neurologic examination was unremarkable except for the altered state of consciousness. The peripheral leukocyte count was $2 \times 10^9/L$ with 6% eosinophils. Chest roentgenogram showed

right paratracheal widening and bilateral proximal upper lobe infiltrates. Cranial computed tomography (CT) showed normal intracranial contents, pansinusitis with bony erosion of the ethmoidal septa and right lamina papyracea, and an ethmoidal sinus mass expanding into the right orbit. Cerebrospinal fluid obtained after the CT contained $5 \times 10^6/L$ leukocytes, 38 mg/dl protein, and 104 mg/dl glucose. The patient was taken to surgery within 12 hours, where decompression of the globe of the eye was achieved by draining a purulent mucocele from the right frontal and ethmoidal sinuses. Bony erosion was found on the floor of the right frontal sinus. The meninges were intact, and there was no cerebrospinal fluid leak. Pathologic examination of the sinus contents revealed cellular debris, eosinophils, Charcot-Leyden crystals, and septate fungi identified as *A. fumigatus* by culture. Administration of amphi-

tericin B was started. On hospital day 8, follow-up cranial CT indicated a new right frontal lobe ring-enhancing lesion. Needle aspiration biopsy revealed *Aspergillus* organisms with an inflammatory reaction composed of eosinophils and Charcot-Leyden crystals (Fig. 1).

Chest CT to evaluate the abnormal chest x-ray film revealed bronchiectasis in the superior segment of the right lower lobe. Fiberoptic bronchoscopy showed an endobronchial mass lesion consistent with granulation tissue occluding the superior segment of the right lower lobe. Culture of bronchial washings grew *A. fumigatus*, and microscopic examination revealed chronic inflammation with eosinophils and hyphae. The skin prick test response to *A. fumigatus* extract was positive. Serologic evaluation revealed a total serum IgE level of 6048 ng/dl (normal value, 1 to 1000) and positive precipitating antibodies to *A. fumigatus*. ELISA for *A. fumigatus*-specific antibody revealed IgE and IgG antibody indices of 5.40 and 15.40, respectively (normal values <2.0). Pulmonary function tests revealed a forced vital capacity (FVC) of 3.70 L (73% of predicted value), forced expiratory volume in 1 second (FEV₁) of 3.13 L (78% of predicted value), and FEV₁/FVC of 85%. No significant change in FEV₁ occurred after administration of inhaled bronchodilator. In order to ascertain whether the patient did indeed have asthma, a methacholine challenge test was performed. The patient was hyper-responsive with a 30% fall in FEV₁ after 7.5 dose units of methacholine. FEV₁ returned rapidly to baseline after administration of inhaled bronchodilator.

The patient underwent excision of the frontal lobe brain abscess on hospital day 29, had an uneventful postoperative course, and was discharged on hospital day 44. He received intravenously administered amphotericin B at home for a total dose of 3 gm. Two months after discharge, he underwent definitive sinus debridement. He was subsequently started on a regimen of intranasal beclomethasone dipropionate and orally administered prednisone, 40 mg, on alternate days. He has done well during 1-year follow-up, and his prednisone dose has been tapered, guided by his IgE level.

DISCUSSION

Fungal sinusitis may manifest in various ways including: (1) fulminant invasive infection, as seen in immunocompromised hosts such as patients with leukemia (*Aspergillus*) and patients with diabetes mellitus (*Mucor*); (2) indolent or chronic invasive infection in normal hosts; (3) mycetoma; and (4) allergic fungal sinusitis, as in AAS. Our patient exhibited the unique pathologic features of AAS as described in 1983 by Katzenstein et al.,² consisting of "allergic mucin" characterized by cellular debris, degenerating eosinophils, Charcot-Leyden crystals, and *Aspergillus* hyphae.

The unusual aspect of this case is that the patient had an *Aspergillus* brain abscess, which was clearly due to his underlying AAS. *Aspergillus* is generally believed to be noninvasive in ABPA and AAS, although a rare case of local bony invasion has been described in a patient with ABPA.³ The mechanism of intracranial spread may have been via the sphenoidal vessels, as noted in nonallergic fungal sinusitis.⁴ We presume that lack of proper sinus drainage with associated increased sinus pressure contributed to development of the brain abscess in this patient. No evidence of contiguous spread to the brain or local bony invasion by fungi was found, nor was there evidence of hematogenous dissemination. Bony erosion has been described previously in AAS but is believed to be secondary to bone remodeling and resorption from pressure mechanisms rather than invasion of fungi.⁵ Proptosis can occur in the presence of a sinus mucocele and is due to expansion of the ethmoidal roof or posterior fossa of the frontal sinus. The intense eosinophilic infiltrate with Charcot-Leyden crystals in the brain abscess suggests that *Aspergillus* antigens can trigger a hypersensitivity reaction in tissue other than bronchi and sinuses in sensitized patients. The role of nasal corticosteroids in delaying or enhancing the progression of this disease is not known.

The concurrence of AAS and ABPA, as seen here, has been described twice previously.^{6, 7} ABPA is characterized by central bronchiectasis, fixed or transient pulmonary infiltrates, peripheral eosinophilia, an elevated *Aspergillus*-specific IgE level, immediate skin reactivity, and precipitating antibodies to *Aspergillus* antigens. It is not clear why AAS and ABPA are not found in association more frequently, in view of their similar immunopathologic responses. Possible explanations include a lack of recognition of sinus disease in patients with ABPA, a need for sinus disease or sinus manipulation for AAS to occur, or the degree of exposure to *Aspergillus*.

In summary, this case demonstrates that AAS can become invasive if untreated and should be considered as part of the differential diagnosis of refractory chronic pansinusitis in young, immunocompetent, atopic patients. Although the ideal therapy is not known, current recommendations include surgical debridement, aeration of the sinuses, and long-term administration of systemic corticosteroids.⁸ Systemic antifungal therapy is indicated only in cases of invasive disease.

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Making maple syrup: Hazardous avocational ingestion of raw sap in a patient with nut and tree pollen sensitivity

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Individuals sensitized to tree pollen may have clinical symptoms ranging from oropharyngeal pruritis to full-blown anaphylaxis on ingestion of the fruit or nuts of northern trees. These reactions are attributable to the presence of various tree pollen-like antigens present in these foods.¹⁻⁵ Some individuals produce tree pollen-specific IgE that recognizes heat-labile antigens. They experience oropharyngeal pruritus when eating fresh, but not cooked or processed (i.e., denatured), fruit. This is a case report of one such patient who had a systemic reaction after ingesting raw maple sap. The case is consistent with an allergic reaction caused by tree pollen-like heat-labile antigens contained in the sap and suggests that raw maple sap should be added to the list of foods

recognized as causing reactions in tree pollen-sensitized individuals.

CASE REPORT

The patient lives in rural northern Ontario and participates in the collection and processing of maple tree sap to make maple syrup in the early spring. On one occasion, she accidentally ingested a few milliliters of raw sap, and within 5 minutes, diffuse erythema, pruritus, and urticaria developed. She had no nasal symptoms, shortness of breath, or wheezing. The eruption lasted for several days, and antihistamines were helpful. She had always tolerated fully processed (i.e., boiled) maple syrup in the past and continues to do so.

The patient has a long history of oropharyngeal pruritus when she eats fresh apples, but she has no symptoms if the apples are cooked. She has both oropharyngeal pruritus and tongue swelling after ingesting walnuts or hazelnuts. There is no history of symptomatic allergic rhinoconjunctivitis during tree pollen season.

Allergy testing was performed (prick method) with standardized extracts (Bencard, Mississauga, Ontario) and appropriate positive and negative controls. She had a 2+ reaction to both tree pollen and hazelnut. Unfortunately, unprocessed maple sap was not available for testing when the patient was first presented. She there-

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