

Bone erosions associated with allergic fungal sinusitis

Jamil N. Al-Swiahb, MD, SSC (ORL),
Surayie H. Al-Dousary, MD, FRCS.

Allergic fungal sinusitis (AFS) is a recently recognized form of benign, non-invasive, fungal sinusitis caused by intense immune reaction to fungal antigens. In 1983, Katzenstein et al¹ reported 7 cases with nasal polyposis, and opacification of multiple sinuses requiring multiple surgeries. They described a histologically distinct mucinous material, containing eosinophils, Charcot-Leyden crystals, and fungal hyphae in tissue resected from the sinuses. In 1994, Bent and Kuhn² published their case series of 15 patients and sought to determine the specific diagnostic criteria for the diagnosis of AFS. Over the past decade, many studies have attempted to elucidate the incidence and clinical course of the disease as well as response to treatment. Bony erosion with subsequent intracranial/intraorbital extension associated with AFS has previously been documented.^{3,4} Its incidence, however, varies from one geographical area to another according to the incidence of AFS.⁴ Allergic fungal sinusitis is common in Saudi Arabia and in this study we evaluate the incidence and characteristics of bone erosions in our population with diagnosis of AFS.

A retrospective chart review was performed in all patients with diagnosis of AFS undergoing endoscopic sinus surgery at King Abdulaziz University Hospital, Riyadh, Saudi Arabia from January 2000 to December 2006. The study was approved by the Rhinology Unit and Rhinology Chair Research Committee of ORL Department of King Saudi University, Riyadh, Saudi Arabia. Eighty-four patients with AFS (presenting at least 4 of Kuhn and Swain AFS diagnostic criteria) met the inclusion criteria. Twenty-two patients, comprising 13 women (59.1%) and 9 men (40.9%), presented with radiographic evidence of skull base erosion or intraorbital extension was included the study. The mean age of the AFS population with bony erosion was 20.6 years (average 8-45). Seven patients (31.8%) had undergone endoscopic sinus surgery without iatrogenic injury to the skull base or orbit before treatment. Fifteen patients presented with unilateral extensive sinus disease, slightly more frequent in the left side 12 (54.5%). Bilateral disease was diagnosed in 7 patients (31.8%). Sites of erosion are shown in Table 1.

Surgical treatment included endoscopic sinus surgery in 11 (50%) patients, computer assisted endoscopic sinus surgery in 10 (45.5%) patients, external approaches in

2 patients (9.1%). Histologic examination revealed chronic inflammatory cell infiltration consisting mostly of eosinophils with septate hyphae that branched at an angle of approximately 45 degrees. These were present in 13 (59.1%) patients. Fungal culture was positive in 8 (36.4%) patients. Five patients (22.7%) grew *Aspergillus species*, 2 patients had *Bipolaris* and one patient grew *Alternaria*. Allergic mucin with dark brown buds was present in 21 patients (95.5%). All patients were followed at least 18 months, in each visit we looked for recurrence of symptoms, and during the endoscopic nasal examination we looked for evidence of recurrence of the obstructive polyposis and postoperative complications. All patients were treated postoperatively with broad spectrum antibiotics, systemic and topical steroids for at least 2 weeks then followed by topical steroids. We observed a recurrence of the AFS in 2 patients (9.1%) after 12 weeks, 8 patients (36.4%) after 24 weeks, and 10 patients (45.5%) after one year. No significant complications were encountered.

Allergic fungal sinusitis is defined by the presence of allergic mucin comprised of necrotic inflammatory cells, Charcot-Leyden crystals, and scattered fungal forms surrounded by an intense mucosal reaction without tissue invasion. The presence of bony erosion of the skull base and orbital in AFS has been well documented in the literature.^{1,2,3,5} Its incidence varies from one geographic area to another and various studies report a range between 20-90%.¹ Previous studies have reported^{3,4} a bone erosion incidence of 25% in children as compared with a 23% in adults. A recent publication by Wise et al³ demonstrated a bone erosion rate of 44.7%. In our study, the incidence of bone erosion was 26%, with a predominance of unilateral disease

Table 1 - Sites of erosion.

Sinuses and subsists	Number of patients (%)	
<i>Ethmoid sinus</i>		
Lamina papyracea	10	(45)
Roof of ethmoid	2	(9.1)
<i>Frontal sinus</i>		
Anterior wall	2	(9.1)
Posterior wall	3	(13.6)
Floor	4	(18.1)
<i>Sphenoid sinus</i>		
Planum sphenoidale	1	(4.5)
Posterior wall	3	(13.6)
Lateral wall	0	
Clivus	1	(4.5)
<i>Maxillary sinus</i>		
Roof	0	
Anterior wall	1	(4.5)
Posterior wall	1	(4.5)
Lateral wall	0	

(54.5%). Allergic fungal sinusitis-related bone erosion most commonly occurred in the ethmoid sinuses followed by sphenoid and frontal sinuses.³ Excluding the medial maxillary sinus wall bone erosion (ostioameatal complex), the lamina papyracea is the most common ethmoid subsite with bone erosion. Henceforth, the orbit was the most common site of extension. This is consistent with the apparent propensity for erosion of the lamina papyracea.³ The anterior cranial fossa was the most common intracranial space affected, although it can affect the middle and posterior cranial fossa, depend on the location of bone erosion as related to the intracranial cavity.³ In our analysis, we found that the ethmoid sinus was the most frequently eroded area (54.5%), followed by the frontal sinus (40.9%), sphenoid sinus (22.7%), and maxillary sinus (9.1%). Previous study reported that lamina papyracea is the most common ethmoid subset with bone erosion,¹² and the orbit is the most common site of extension (36.3%) and intracranial extension in 7 patients (31.8%). Alteration in the sinus bone in AFS patients is thought to result largely from pressure remodeling (from a combination of the underlying sinonasal polyposis and the allergic mucin) rather than overt bone destruction. Others have suggested that this process may aid by the inflammatory mediators released from eosinophils.¹⁻³ Moreover, others suggest progressive bone demineralization of the sinus walls by the allergic mucin alone or by the underlying polyposis.^{1,2} Many diseases can cause bone erosion, such as chronic rhinosinusitis, mucocele, and neoplasm. If these are excluded, AFS is the most common disease causing bone erosion.^{1,4} Many authors stressed that AFS is usually non-invasive because the hyphal fragments did not invade. Bony erosion, is common in AFS, however, deeper invasion into the underlying bone is very unusual. To our knowledge, histological findings of bone invasion and necrosis present in invasive form have not been described in allergic fungal sinusitis.^{1,3}

To date, no one has examined the role of socioeconomic factors and/or certain geographic and environmental factors in AFS in the development of sinus bone erosion. Gupta et al⁴ reported an incidence of bony erosion in 88% of children compared to 36% in adults. Wise et al³ suggested that bony erosion is more common in African-Americans.

Computerized tomography scan findings may be confused with those of a malignancy and include complete opacification of at least one of the involved sinuses; increased intrasinus attenuation, expansion of an involved sinus, remodeling and thinning of

the bony sinus walls, and erosion of the sinus wall. Multisinus involvement may be unilateral or bilateral.^{1,3} Computerized tomography cannot distinguish between bone erosion caused by direct hyphal invasion and pressure erosion. Allergic fungal sinusitis is characterized by a special type of mucin, termed "allergic mucin", which is present in variable amounts. It should be sought for in every specimen and submitted for microscopic examination and culture.

Fungi that are most often identified in AFS are dematiaceous types, but *Aspergillus* is most commonly isolated in mycetoma.^{1,4} However, there is some variation in the types of fungi isolated in AFS; in some geographic areas, *Aspergillus species* are the most common.⁴ Indeed, in our area and studies *Aspergillus species* was the most common organism. There is no clear evidence in the literature on the relation of specific fungal organism to bone erosion especially in AFS; however, we found that most culture positive AFS in our area revealed *Aspergillus species* especially if the disease was associated with bone erosion. Current treatment of allergic fungal sinusitis consists of a combination of sinus surgery and corticosteroid therapy, complete removal of fungi and allergic mucin from the sinus through an endoscopic approach is the current cornerstone of therapy. Advancement in endoscopic sinus surgery and development of computer-aided (image-guided) endoscopic sinus surgery will lead to increase surgical effectiveness and decrease surgical morbidity. Concomitant treatment with systemic corticosteroids provides the best long-term relief of symptoms, but the exact dosing regimen and duration of therapy have yet to be established. Some doubt remains as to whether a long-term cure for this condition exists. Systemic antifungal agents have not shown any effectiveness in allergic fungal sinusitis and should be avoided. In this diagnostic and therapeutic dilemma, long-term follow up is recommended to decrease the recurrent rate. All our patients underwent powered endoscopic sinus surgery and we use computer-aided endoscopic sinus surgery in many of our patients and we found it useful to localize the areas of erosion and complete disease eradication. All our patients received systemic and local corticosteroid, and duration and frequency of postoperative visits were determined by patient symptoms and endoscopic sinus evaluation. All patients were followed with recurrent endoscopic examination and CT scans in a period of 1-2 years, with evidence of recurrent disease in 5 (22.7%) of patients not respond to medical treatment and they need revision surgery.

We believe that these immunocompetent patients with erosive fungal sinusitis were not sufficiently

studied and the disease origin and pathogenesis is poorly understood. Further studies may be valuable in the assessment of therapeutic efficacy. Even though we found that these patients could be successfully managed with aggressive medical therapy and endoscopic sinus surgery with no serious complications.

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From the Department of Otorhinolaryngology, Head & Neck Surgery, King Abdulaziz University Hospital, Riyadh, Kingdom of Saudi Arabia. Address correspondence and reprints request to: Dr. Jamil N. Al-Swiahb, Department of Otorhinolaryngology, Head & Neck Surgery, King Abdulaziz University Hospital, PO Box 245, Riyadh 11411, Kingdom of Saudi Arabia. Tel. +966 (1) 2355769. Fax. +966 (1) 2350220. E-mail: fawaz8026@yahoo.com

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Related topics

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