Allergic Fungal Rhinosinusitis: Pattern of Orbital Invasion and Incidence of Bone Erosion and Sinus Expansion

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Objective: Allergic fungal rhinosinusitis is a fungal infection of paranasal sinuses that often associated with bone destruction with extension outside the sinonasal framework. The objective of this report is to further define these findings as they relate to this disease.

Study design: This is a prospective non-randomized case series study. One hundred twenty-three patients with allergic fungal rhinosinusitis were enrolled in this study. Radiological evaluation for every patient was done in the form of computed tomography and\or magnetic resonance image in order to define bone erosion and sinus expansion associated with this type of rhinosinusitis.

Results: One hundred and twenty-three cases of proven allergic fungal sinusitis were evaluated. The mean age of the study group was 35.4 and the SD 13.1. The age range was 13 to 61 years. Males comprised 68.3 % of the patients and females 31.7 %. Among 123 enrolled patients, 57.9% of study group showed affection with 54.3% had bone erosion, 21.4% had expansion and 24.3% had both erosion and expansion lesion, 80% of them showed the lesion unilaterally versus 20% had bilateral affection.

Conclusion: Bone invasion is a characteristic feature of allergic fungal sinusitis. Orbital and skull base invasion in these cases is not an uncommon finding. Bone invasion should be addressed during evaluation of every case of allergic fungal sinusitis. According to the results of this study, orbital invasion mostly occurs through the medial wall, followed by the roof of the orbit, and lastly through the floor.
Keywords: Fungal rhinosinusitis, bone invasion, sinus expansion, skull base invasion, orbital invasion.

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Introduction

Allergic fungal rhinosinusitis is a fungal infection of paranasal sinuses that often associated with bone destruction with extension outside the sinonasal framework. [1] Fungal sinusitis is demonstrated radiologically as a nodular inflammation of the mucoperiosteal lining leading to mottling opacification of the cavity of the sinus. In computed tomography, fungal disease is descried as a rim of attenuated soft-tissue of variable thickness along the bony walls of the paranasal sinuses. [2] Five basic types of fungal rhinosinusitis disorder are well known, characterized histopathologically and by different clinical presentation. Three types of fungal rhinosinusitis are characterized by tissue-invasion, acute fulminating (acute invasive) fungal rhinosinusitis, chronic invasive fungal rhinosinusitis and granulomatous invasive (indolent) fungal rhinosinusitis. The non-invasive fungal infections are allergic fungal rhinosinusitis and the fungal ball (sinus mycetoma). [3] Allergic fungal sinusitis (AFS) typically presents a non-aggressive slow course and does not demonstrate tissue-invasion, although sinus expansion and bone erosion of the sinus walls are not uncommon association. Kinsella et al. have suggested a new entity, 'skull base allergic fungal sinusitis', which included the histological features of allergic fungal sinusitis and bone erosion on computed tomography (CT). [4]

Bone erosion associated with AFS can lead to skull base and ophthalmic complications with significant morbidity and possibility of mortality. [5] Ophthalmic manifestations or complications of AFS range from erosion of the orbital medial wall proved on imaging to proptosis and loss of vision in extreme cases. The proximity of critical structures such as the internal carotid artery, cavernous sinus, cranial nerves, dura matter and cerebral lobes makes any or all of these at risk due to the disease progress itself and in surgical treatment as well. Extensive AFS cases with massive skull base erosion and ophthalmic complications are still relatively rare entity and present a diagnostic and therapeutic challenge, often simulating invasive fungal disease or malignancy. [6]

The goal of surgical management of the cases of AFS is to provide adequate sinus aeration and removal of fungal debris. Endoscopic sinus techniques are regarded as effective as the traditional open surgical techniques in treating cases of AFS even in the presence of massive skull base erosion. [7]

Allergic fungal rhinosinusitis is considered as hypersensitivity to extra mucosal fungal antigens, with consequent production and accumulation of allergic mucin. Allergic fungal rhinosinusitis is usually presenting in the form of gradually increasing nasal obstruction, viscous rhinorrhea, hyposmia, nasal crusting. [8]

AFS is characterized by multiple nasal polyps, with unilateral distribution more common than bilateral. The classic diagnostic criteria for AFS were described by Bent and Kuhn, who proposed the following: type 1 hypersensitivity; characteristic computed tomography (CT) scan findings;

eosinophilic mucus, and nasal polyposis without fungal invasion; and a positive fungal stain of the sinus contents. [9]

The purpose of this study is to investigate the incidence of erosion and bone invasion of the orbit and the skull base and the sinus expansion in cases of allergic fungal sinusitis.

Methodology

One hundred and twenty-three patients were enrolled in this study. All of these patients were collected from those attending the out-patient clinic of Otolaryngology department at Fayum university and in the private practice in a period of sixty-one months; from August 2012 through September 2017.

Only patients fulfilling the criteria for diagnosis of the allergic fungal sinusitis according to Bent and Kuhn were enrolled in this study.

Each patient was subjected to history taking, endoscopic nasal examination and CT scan. MRI was performed for those with suspected skull base invasion to evaluate intracranial extension, dural invasion, cavernous sinus or internal carotid artery involvement. The CT and MRI studies were analyzed by a senior radiologist for specific evaluation of bone or orbital invasion in addition to the presence of associated sinus expansion.

All patients underwent endoscopic sinus surgery. Systemic and topical steroids were used in postoperative period. Patients were followed up post-operatively for disease recurrence, sinus patency and mucosal changes for 1 to 3 years. Radiological analysis for the orbit, skull base, frontal, ethmoid, maxillary and sphenoid sinuses was performed regarding bone invasion and sinus expansion as follows:

- Orbital wall invasion; medial (lamina papyracea) wall, roof, floor of the orbit.
- Frontal sinus; floor invasion, anterior table invasion, posterior wall invasion, expansion of the sinus.
- Maxillary sinus; invasion of the medial wall, posterior wall invasion, roof invasion, or sinus expansion.
- Anterior ethmoid group; expansion of the sinus group
- Posterior sinus group; expansion, invasion into the sphenoid sinus or the orbit
- Sphenoid sinus; expansion of the sinus, planum or clival invasion
- Skull base invasion; fovea ethmoidalis, cribriform plate, lateral lamella, cavernous sinus invasion.

The results of radiological analysis were reviewed for each patient. Statistical analysis with a binomial logistic regression (SPSSS 10.0 for Windows, Chicago, IL) was performed on the data with respect to the presence or absence of bone erosion, and for the presence of bone erosion with or without expansile sinus changes.

Results

One hundred and twenty-three cases of proven allergic fungal sinusitis were evaluated. The mean age of the study group was 35.4 and the SD 13.1. The age range was 13 to 61 years. Males comprised 68.3 % of the patients and females 31.7%; the male to female ratio was 2.7:1. The majority of patients (67.32%) were of lower socioeconomic status.

CT paranasal sinus, coronal and axial examination was done for all included patients. These radiological studies were evaluated by a senior radiologist for detection of bone erosion, sinus expansion and extra-sinonasal involvement by the fungal lesion.

Data were collected and coded to facilitate data manipulation and double entered into Microsoft Access and data analysis was performed using Statistical Package of Social Science (SPSS) software version 18 in windows 7. Simple descriptive analysis in the form of numbers and percentages for qualitative data, and arithmetic means as central tendency measurement, standard deviations as measure of dispersion for quantitative parametric data. One way ANOVA test in comparing more than two independent groups of quantitative data. Chi square test to compare two of more than two qualitative groups. The P-value ≤ 0.05 was considered the cut-off value for significance.

Among 123 enrolled patients, 57.9% of study group showed affection with 54.3% had bone erosion, 21.4% had expansion and 24.3% had both erosion and expansion lesion, 80% of them showed the lesion unilaterally versus 20% had bilateral affection.

The most common affected site was the orbit (18.6%), followed by the sphenoid sinus (17.1%) then the anterior ethmoid group (15.7) as shown in **Table 1**.

Table 1. Frequency of the lesion character among study group.

Variables	Number (n=123)	%
Lesion		
No	58	47.2%
Yes	65	52.8%
Type of lesion (n=65)		
Erosion	32	49.2%
Expansion	13	20%
Both	20	30.8%
Side of lesion		
Unilateral	51	78.5%
Bilateral	14	21.5%
Affected sinus		
Frontal	6	9.1%
Maxilla	6	9.1%
Anterior ethmoid	11	16.7%
Posterior ethmoid	6	9.1%
Sphenoid	12	18.2%
Orbital	10	15.2%
Anterior skull base	7	10.6%
Middle fossa	3	4.5%
Posterior ethmoid& Orbital	2	3%
Frontal& Anterior skull base	2	3%
Frontal& Orbital	1	1.5%

There was statistically significant difference with p-value <0.05 in the type of the lesion (erosion, sinus expansion or both) and the site of affection (orbit, sinus or extrasinonasal site) as shown in **Table 2**.

Table 2. Comparison of lesion types in different sites.

Variables	Type of lesion			p-value
	Erosion	Expansion	Both	
	No. (%)	No. (%)	No. (%)	
Frontal	2 (33.3%)	1 (16.7%)	3 (50%)	
Maxilla	1 (16.7%)	2 (33.3%)	3 (50%)	
Anterior ethmoid	5 (50%)	1 (10%)	4 (40%)	
Posterior ethmoid	-----	4 (66.7%)	2 (33.3%)	
Sphenoid	3 (25%)	5 (41.7%)	4 (33.3%)	
Orbital	10 (100%)	-----	-----	0.001*
Anterior skull base	7 (100%)	-----	-----	
Middle fossa	3 (100%)	-----	-----	
Posterior ethmoid& Orbital	-----	-----	2 (100%)	
Frontal& Anterior skull base	1 (50%)	-----	1 (50%)	
Frontal& Orbital	-----	-----	1 (100%)	

No posterior fossa involvement was detected among the studied patients.

There was statistically significant difference with p-value <0.05 between both sexes and the type of the lesion with high percentage of males (65.2%) showed erosion but 37.5% of females had both erosion and expansion.

On the other hand, there was no statistically significant difference with p-value <0.05 as regards age and side of the lesion as shown in **Table 3**.

Table 3. Comparison of the lesion types and the sex and side of the lesion.

Variables	Type of lesion			p-value
	Erosion	Expansion	Both	
Age	37.9±13.9	34±12.4	36.9±13.3	0.7
Sex				
Male	26(60.5%)	7(16.3%)	10(23.3%)	0.03*
Female	6(27.3%)	6(27.3%)	10(45.5%)	
Side of lesion				
Unilateral	25(49%)	12(23.5%)	14(27.5%)	0.3
Bilateral	7(50%)	1(7.1%)	6(42.9%)	

Orbital invasion was characterized by the following:
Lamina papyracea was the most common site to be affected. However, the invasion of the anterior part of the lamina (anterior to the level of the posterior wall of maxilla) was invaded more (8 out of 13 cases) than the posterior part through the posterior ethmoid sinuses (5out of 13 cases).

The roof of the orbit was the second site to be affected. This invasion occurred either through the medial portion of the roof which was more common (7 out of 9 cases) or through the lateral part of the roof (2 out of 9 cases).

The floor of the orbit was the last site in incidence to be invaded.

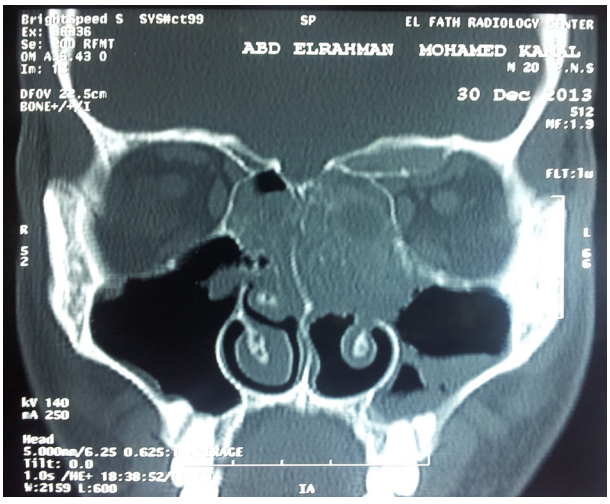

Fig 1. Non-enhanced coronal CT demonstrating AFS with roof and medial wall invasion of the Lt. orbit.

Discussion

AFS appeared as a distinct clinical entity in the early 19808. The affected individuals are immunocompetent with a history of chronic bacterial or polypoid sinusitis. Goldstein et al5, reported among AFS affected individuals a history of atopy in one third of patients, and a history of asthma in another one third. The disease can be suggested preoperatively from the typical CT and MRI features, but it must always be confirmed histologically.

Fig 2. Non-enhanced axial cut CT demonstrating AFS with massive medial orbital invasion.

Zinreich et al, [9] described areas of increased attenuation inside the sinus cavity in CT scans without contrast in patients with fungal sinusitis.

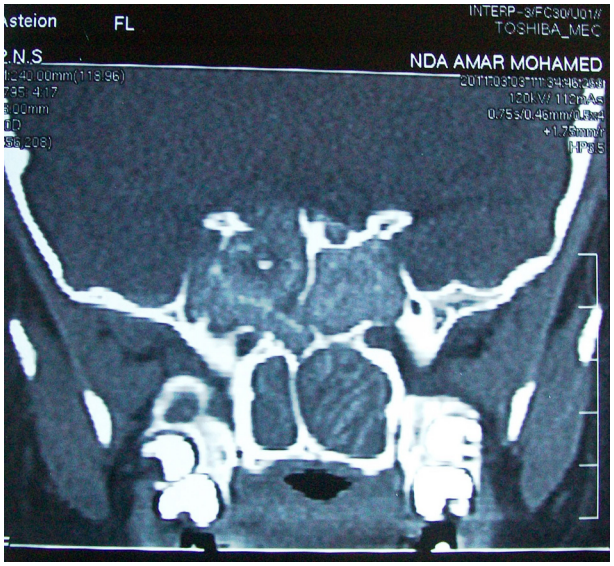

Fig 3. Non-enhanced coronal cut CT of a case of AFS demonstrating bone erosion of the posterlateral wall of the sphenoid and the clivus.

It is well known that; bone erosion is a common finding among cases of AFS. Studies in this field has been reported the incidence of bone erosion for patients with AFS to be in the range of 20% to 90%. [10]

AFS-related bone erosion can be attributed to the pressure atrophy from expansile growth of polyps or mucoceles. Inflammatory mediators produced in association with allergic mucin have also been implicated. [11] Direct evidence supporting either hypothesis remains scant. Clearly, bone erosion has also been described in association with long-standing expansile processes, such as sinonasal polyposis or mucoceles not related to AFS. Handley et al [12] reported that bone fragments from AFS-related bone erosion histologically demonstrated changes compatible with an active remodeling process. Sinus expansion is not uncommon in cases of AFS. It is postulated that ubiquitous fungi become deposited in the sinus and produce Type I (IgE)– and Type III (immune complex)–mediated reactions that trigger an intense eosinophilic inflammatory response. The inflammation leads to sinus obstruction followed by stasis and then accumulation of allergic mucin. This environment is ideal for proliferation of the fungus with subsequent antigenic exposure and a host response, leading to nasal obstruction and allergic mucin. A vicious circle of increased sinus mucosal inflammation with sinus obstruction, less ventilation, and further more antigen contact. Within an involved paranasal sinus cavity, the allergic fungal mucin accumulates and begins to resemble and behave similarly to a mucocele. As the "fungocele" expands with time, bone erosion and sinus expansion both can cause intracranial or intraorbital extension without tissue invasion. [6]

Some studies demonstrated that, this expanding mass leads to bony erosion and involvement of adjacent structures in 6–56% of AFS patients. [13]

This study included 123 patients with AFS. 57.9% of the study group showed affection in sinuses with 54.3% had bone erosion, 21.4% had expansion and 24.3% had both erosion and expansion lesion, 80% of them showed lesion unilaterally versus 20% had bilateral affection.

Liu et al [1] described 38% of their study group with AFS to have erosion of bone. On the other hand, Kinsella et al [4] described only 21.4%. Nussenbaum et al. [14] documented 20% of cases of their study to have bony invasion. Qazi et al. [15] presented 39% of their studied cases with orbital signs and symptoms to have AFS. Marfani et al16 presented 30.04% of their studied AFS have combined orbital and cranial invasion.

This study demonstrated that, males showed more incidence of bone erosion than females. This can be explained on the basis that, males usually seek medical advice than females and at the same time, in these low socio-economic class of patients the presentation for management is usually late.

On the other hand, females, according to this study results, showed more incidence of sinus expansion than males. This can be attributed to the fact that, bone density in females is less than those of males leading to easy ballooning of the affected sinus than in males.

The orbital invasion is an important item during evaluation of the cases of AFS. This invasion may be presented clinically or by radiological investigation. The most common site of bone invasion in the orbit is the medial wall, followed by the roof, followed by the floor according to the results of this study.

Sinus expansion is a sinus ballooning due to accumulation of the expanding mucin before invasion of the sinus bony wall. The clinical significance of this radiological finding is to predict disorientation and anatomical difficulties and during endoscopic management of these cases.

Conclusion

Bone invasion is a characteristic feature of allergic fungal sinusitis. Orbital and skull base invasion in these cases is not

an uncommon finding. Bone invasion should be addressed during evaluation of every case of allergic fungal sinusitis. According to the results of this study, orbital invasion mostly occurs through the medial wall, followed by the roof of the orbit, and lastly through the floor. Sinus expansion associated with these cases should be expected to deal with during the endoscopic management.

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