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Clinical presentation of allergic fungal sinusitis in Aden, Yemen

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Abstract: Allergic fungal sinusitis, or eosinophilic fungal rhinosinusitis, is a barely recognized pathologic entity that belongs to the fungal rhinosinusitis group. It is characterized by pathognomonic eosinophilic mucin-containing hyphae besides other distinctive histological and imaging features that contribute to the diagnosis. The aim of the study was to assess the presentation of allergic fungal sinusitis and to evaluate the different clinical features of this clinical dilemma in Aden, Yemen. We performed a retrospective chart review of 64 patients with allergic fungal sinusitis. They were (64.1%) males and (35.9%) females with ratio male to female 1.8:1. Their mean age was (25.9 ± 9.02) years, range (10–45) years. The most common symptoms was nasal obstruction 64 (100%) followed by nasal discharges 35 (54.7%), headache 17 (26.6%), and anosmia 16 (25%). The difference between values of nasal discharge is statistically significant ($p < 0.05$). Nasal polyps is the most common signs 64 (100%) followed by facial disfigurement and ocular abnormalities (OA) 19 (29.7%). By CT-Scan examination, the nasal polyps were 49 (76.6%) unilateral side and 15 (23.4%) bilateral side. Sinus expansion and double sign density found in all patients 64 (100%) The difference between values of nasal discharge is statistically significant ($p < 0.05$). Total serum IgE ranged between 500 IU/ml and 1200 IU/ml. The mean value of serum IgE of all patients was 704.9 ± 127.7 IU/ml and in male patients was 682.6 ± 94.9 IU/ml while in female patients was higher 744.9 ± 166.6 IU/ml. Polyps, nasal discharge, facial disfigurement & ocular abnormalities, orbital extension, headache, anosmia and skull base erosion were the most common presentations. Males are most commonly affected and most commonly affected age group is ≤ 20 to 30 years.

Key words: Clinical presentation; allergic fungal; rhinosinusitis; Aden; Yemen

Introduction

Allergic fungal sinusitis (AFS), or eosinophilic fungal rhinosinusitis, is a barely recognized pathologic entity that belongs to the fungal rhinosinusitis group [6]. It is broadly defined as a non-invasive fungal infection of sinuses inducing a marked type I hypersensitivity reaction that overshadows the clinical picture [7,15]. It is characterized by pathognomonic eosinophilic mucin-containing hyphae besides other distinctive histological and imaging features that contribute to the diagnosis [2,10,18,20].

The disease was first recognized in 1976 when Safirstein published the first description of Allergic Fungal Rhinosinusitis (AFRS) [23].

He observed the clinical similarity that this assemblage of findings shared with allergic bronchopulmonary Aspergillosis [17, 23]. Katzenstein, et al. later on further described 7 similar cases, which was termed as allergic aspergillus sinusitis [11]. It is interesting to find that several reports in the available literature have described AFRS with different clinical presentations; some even involving more than one type of fungal rhinosinusitis occurring concurrently [22, 30].

The aim of the study was to assess the presentation of allergic fungal sinusitis and to evaluate the different clinical features of this clinical dilemma in Aden, Yemen

Materials and method:

Study design, place and time: We performed a retrospective chart review of all patients presenting to the

ENT outpatient clinic at Algamhoria Teaching Hospital over a 2-year period from January 2018 to December 2019.

Number and assessment of cases: Sixty-four patients were diagnosed with allergic fungal sinusitis using Bent-Kuhn criteria [5] through history, physical examination, endoscopic examination, serology and CT-scan of the nose and paranasal sinuses.

Data collection: All information was obtained from handwritten paper charts used in the ENT clinic. The collected data were sex, age, clinical presentation, CT-Scan examination and laboratory results.

Data analysis: The collected data were tabulated and statistical analysis was done by estimating rates, means and

standard deviations, Fisher test was used and p-value < 0.05 was considered as statistically significant. The statistical software package SPSS version 17 was used.

Results:

During the two years study period, 64 patients presented with allergic fungal sinusitis and seen at our private clinic. There were 41 (64.1%) males and 23 (35.9%) females with ratio male to female 1.8:1. The mean age of all patients was (25.9 ± 9.02) years, range (10– 45) years, (27.5 ± 8.9) for males, and (23.1 ± 8.8) years for females, (P > 0.05), all variables illustrated in Table 1 and Figure 1.

Table 1: Distribution of demographic characteristics of the study patients (n = 64)

Variable	Ratio	Range	Mean	No	%	p-value
Sex:						
Males				41	64.1	
Females				23	35.9	
Male to Female	1.8 : 1					
Age range (years):		10 – 45				
Mean age ± SD* (years):						
All patients			25.9 ± 9.02			P > 0.05
Male patients			27.5 ± 8.9			
Female patients			23.1 ± 8.8			
Age groups (years):						
≤ 20				21	32.8	
21 – 30				21	32.8	
> 30				22	34.4	

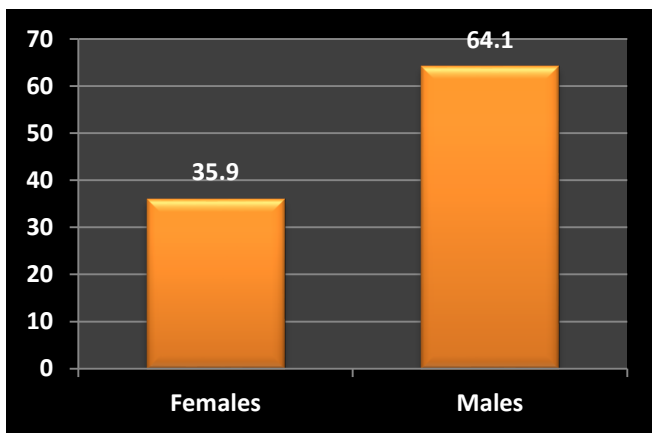


Figure 1: Distribution of patients related to sex

Table 2 reveals the distribution of clinical features, CT-Scan findings and mean values of serum IgE among the study patients related to sex.

The most common symptoms was nasal obstruction 64 (100%) followed by nasal discharges 35 (54.7%), headache 17 (26.6%), and anosmia 16 (25%).

The difference between values of nasal discharge is statistically significant (p < 0.05).

Nasal polyps is the most common signs 64 (100%) followed by facial disfigurement and ocular abnormalities (OA) 19 (29.7%).

By CT-Scan examination, the nasal polyps were 49 (76.6%) unilateral side and 15 (23.4%) bilateral side, as shown also in Figures 2.

Sinus expansion and double sign density found in all patients 64 (100%) also, orbital extension 19 (29.7%) and skull base erosion 16 (25%).

Total serum IgE ranged between 500 IU/ml and 1200 IU/ml. The mean value of serum IgE of all patients was 704.9 ± 127.7 IU/ml and in male patients was 682.6 ± 94.9 IU/ml while in female patients was higher 744.9±166.6 IU/ml.

Table 2: Distribution of clinical features, CT-Scan finding and mean values of serum IgE related to sex (n = 64)

Variables	Sex		Total		p-value
	Female No (%)	Male No (%)	No (%)	(%)	
Nasal obstruction: Present	23 (35.9)	41 (64.1)	64 (100)		
Nasal discharge: Yes	7 (10.9)	28 (43.8)	35 (54.7)		P = 0.004
No	16 (25.0)	13 (20.3)	29 (45.3)		
Headache: Yes	5 (7.8)	12 (18.8)	17 (26.6)		P > 0.05
No	18 (28.1)	29 (45.3)	47 (73.4)		
Anosmia: Yes	6 (9.4)	10 (15.6)	16 (25.0)		P > 0.05
No	17 (26.6)	31 (48.4)	48 (75.0)		
Nasal polyp: Present	23 (35.9)	41 (64.1)	64 (100)		
FD*,OA** : Present	9 (14.1)	10 (15.6)	19 (29.7)		P > 0.05
Absent	14 (21.9)	31 (48.4)	45 (70.3)		
Polyp in Sinus (CT-Scan) Bilateral	8 (12.5)	7 (10.9)	15 (23.4)		P > 0.05
Unilateral	15 (23.4)	34 (53.1)	49 (76.6)		
S exp ds density*** (in CT-Scan): Present	23 (35.9)	41 (64.1)	64 (100)		
Orbital extension: Present	9 (14.1)	10 (15.6)	19 (29.7)		P > 0.05
Absent	14 (21.9)	31 (48.4)	45 (70.3)		
Skull base erosion: Present	5 (7.8)	11 (17.2)	16 (25.0)		P > 0.05
Absent	18 (28.1)	30 (46.9)	48 (75.0)		
Serum IgE: Mean value (IU/ml)	744.9±166.6	682.6 ± 94.9	704.9 ± 127.7		P > 0.05

* FD = Facial disfigurement, **OA = Ocular Abnormalities;

*** S exp ds density = Sinus expansion and double sign density;

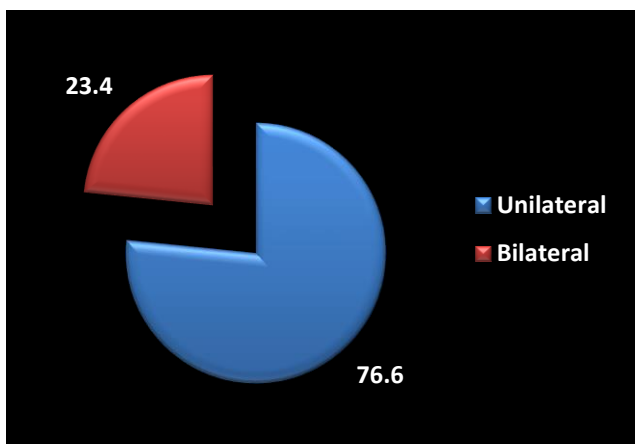


Figure 2: Sinus side involvement in CT-scan of nasal polyps

Discussion:

In this retrospective chart review, we found 64 patients diagnosed with AFS. They were 41(64.1%) males and 23(35.9%) females with ratio male to female 1.8:1.

The findings in the present study were similar to studies reported by Thahim et al [28] and Deshazo et al [8] who found males predominance in their studies.

Conversely Manning et al [16] and Zakirullah et al [29] found female to male ratio predominance.

The mean age of all patients was (25.9 ± 9.02) years, range (10– 45) years, (27.5 ± 8.9) years for males, and (23.1 ± 8.8) years for females, (P > 0.05).

This is quite similar to a study conducted in India in which the mean age was 28.4 years with a range of 18–48 years [12] and two studies in Saudi Arabia by

Al-Mulhem et al [3] with mean age 23 years (age range 7 to 68 years) and the other by Alghonaim et al [1] in which the mean age was 31.57 years with a range of 13–55 years.

However, the current study reveals that all patients were 32.8% aged 20 years aged and less and 32.8% between 21 to 30 years old, so, 65.6% were in 2nd and 3rd decade of

life, which is similar to studies reported in the published literature [8,25,29].

The most common symptoms was nasal obstruction 64(100%) followed by nasal discharges 35 (54.7%), headache 17 (26.6%), and anosmia 16 (25%).

The difference between values of nasal discharge is statistically significant ($p < 0.05$).

Khan et al [13] found in their study headache was in (43.5%) patients.

Zakirullah et al [29] mentioned that nasal obstruction, nasal discharge, symptoms of allergic rhinitis or sinusitis and headache were the main presentation in their patients.

The clinical findings in published literature are more or less the same with insignificant difference in frequencies of the symptoms [19,29].

Nasal polyps is the most common signs 64 (100%) followed by facial disfigurement and ocular abnormalities (OA) 19 (29.7%).

The finding of the present study is comparable with findings reported by Awan et al [4] and Khattar et al [14].

In a study conducted by Kaur et al [12] polyps were seen in 95% of patients with AFRS. Slavin [27] reported that patients with allergic fungal rhinosinusitis present with symptoms of chronic rhinosinusitis to conventional medical therapy, nearly always with concomitant nasal polyps.

Santhi et al [24] found in their study 33.3% were with symptoms of nasal polyposis. Also, in a study conducted by Shah et al, 49% of patients came with signs of nasal polyps [26].

By CT-Scan examination in the present study there were (76.6%) unilateral side and bilateral sides (23.4%).

Mukherji et al [21] reported in their study finding that, approximately half the cases (51%) occur unilaterally.

The present study showed that skull base erosion was (25%). Several studies have quoted the incidence of bony erosion with spread of pathology into the adjacent anatomic areas as 20% [21].

The present study revealed that the total serum IgE ranged between 500 IU/ml and 1200 IU/ml. The mean value of serum IgE of all patients was 704.9 ± 127.7 IU/ml and in male patients was 682.6 ± 94.9 IU/ml while in female patients was higher 744.9 ± 166.6 IU/ml.

The present study results are in agreement with Ferguson [9] who stated that total IgE in patients with AFS is frequently elevated with a mean of 668 IU/mL

Conclusion:

Allergic fungal rhinosinusitis can present clinically in different ways. Despite being categorized as a benign, non-invasive disease, its presentation can range from nasal obstruction to signs and symptoms of intraorbital and/or intracranial complications.

The clinical presentation of allergic fungal sinusitis not only confined to nose and paranasal sinuses but may be extended to surrounding structures, so cooperation between ENT specialist and ophthalmologist, neurosurgeon and radiologist should be present.

The hot climate in Aden and south governorates with present of nasal atopy lead to prevalence of this clinical entity.

Understanding of these clinical presentations prevent the delayed presentation of patients to ENT specialist, so lead to early diagnosis and prevent the complications of this clinical dilemma.

Nasal obstruction, nasal discharge, headache, anosmia, facial disfigurement & ocular abnormalities, orbital extension, and skull base erosion with and without intracranial extension were the most common presentations. Males are most commonly affected and most commonly affected age group is ≤ 20 to 30 years.

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المظاهر السريرية لالتهاب الجيوب الأنفية الفطري التحسسي في عدن، اليمن

صالح محمد أبوبكر الشعبي¹

الملخص: التهاب الجيوب الأنفية الفطري التحسسي، أو التهاب الجيوب الأنفية الفطري البيوزيني، هو كيان مرضي لا يكاد يعرف وينتمي إلى مجموعة التهاب الجيوب الأنفية الفطرية. يتميز بخيوط خيطية محتوية على البيوزينيات مرضية إلى جانب ميزات نسجية وتصويرية مميزة أخرى تسهم في التشخيص. كان الهدف من الدراسة هو تقويم مظاهر التهاب الجيوب الأنفية الفطري التحسسي وتقويم السمات السريرية المختلفة لهذه المعضلة الطبية في عدن، اليمن. أجرينا مراجعة للملفات الطبية بأثر رجعي لـ 64 مريضاً يعانون من التهاب الجيوب الأنفية الفطري التحسسي. كانوا (64.1%) ذكوراً و (35.9%) إناثاً بنسبة ذكور إلى إناث 1.8:1. كان متوسط أعمارهم (9.02 ± 25.9) سنة، المدى (10-45) سنة. كانت الأعراض السريرية الأكثر شيوعاً هي السلائل الأنفية 64 (100%) والانسدادات الأنفية 64 (100%)، تليها إفرازات الأنف 35 (54.7%)، وتشوه الوجه مع تشوهات في العين والامتداد المداري لكل منها بنسبة 19 (29.7%)، والصداع 17 (26.6%)، وفقدان حاسة الشم وتآكل قاعدة الجمجمة كل واحد بنسبة 16 (25%). بوساطة الأشعة المقطعية وجدت السلائل الأنفية 49 (76.6%) جانب أحادي وثنائي الجانب 15 (23.4%). تم العثور على تمدد الجيوب الأنفية وكثافة الإشارة المزدوجة في جميع المرضى (100%) الفرق بين قيم إفرازات الأنف ذو دلالة إحصائية (P < 0.05). تراوح إجمالي مصل الدم IgE بين 500 وحدة دولية / مل و 1200 وحدة دولية / مل. كان متوسط قيمة مصل IgE لجميع المرضى 704.9 ± 127.7 وحدة دولية / مل وفي المرضى الذكور كان 94.9 ± 682.6 وحدة دولية / مل بينما كان لدى المرضى الإناث 166.6 ± 744.9 وحدة دولية / مل. كانت أكثر أعراض شيوعاً هي السلائل الأنفية، وإفرازات الأنف، وتشوه الوجه، وتشوهات العين، وتمدد الحجاج، والصداع، وفقدان الشم، وتآكل قاعدة الجمجمة وكان الذكور هم الأكثر شيوعاً كما وجد أن الأكثر شيوعاً هم الفئة العمرية المتأثرة ≥ 20 إلى 30 عاماً.

الكلمات المفتاحية: مظاهر سريرية، حساسية فطرية، التهاب الجيوب الأنفية، عدن، اليمن