

Sinonasal Anatomic Variants in Allergic Fungal Rhinosinusitis

Chadi A. Makary, MD¹ , Brock Parman, BS², Brittany Gill, BS², Aykut Unsal, DO³ , Thomas Holmes, MD², Camilo Reyes, MD², and Stilianos E. Kountakis, MD, PhD²

Abstract

Background: The pathophysiology of allergic fungal rhinosinusitis (AFRS) is not well understood.

Objectives: To study the incidence of sinonasal anatomic variants (AVs) in AFRS.

Methods: Retrospective cohort study. Patients with AFRS presenting to our clinic from 2008 to 2018 were reviewed for laterality of the disease. Patients with unilateral involvement were further studied for AV presence by reviewing their preoperative sinus CT scan at original presentation. Each patient's uninvolved side served as its own control. AVs studied include agger nasi (AN), Haller's cells (HC), concha bullosa (CB) and supraorbital ethmoid cells (SOEC).

Results: A total of 155 patients with AFRS were identified. 30 patients (19.3%) had unilateral disease involvement. Of these, 16 patients had left sided and 14 patients had right sided disease. AV were present in 26 of the 30 AFRS sides (86.7%) and in 21 of the 30 healthy sides (70%), (chi squared = 2.45, $p = 0.117$). CB were present in 12 of 30 AFRS sides (40%) and only in 1 of 30 healthy sides (3%), (chi squared = 11.88, $p = 0.0006$). The presence of HC was more common in the AFRS vs healthy sides (7 of 30, 23.3% vs 2 of 30, 6.7%, respectively, $p = 0.071$). AN and SOEC presence was similar in both AFRS and healthy sides ($p = 0.598$ and $p = 0.718$ respectively).

Conclusion: AV are more common on the sides with AFRS compared to healthy sides, with the presence of CB reaching unquestionable statistical significance. Further study is needed to determine the possible association of AV, especially concha bullosa, with the pathophysiology of AFRS.

Keywords

allergic fungal rhinosinusitis, anatomic abnormalities, concha bullosa

Introduction

Allergic fungal rhinosinusitis (AFRS) is a severe form of chronic rhinosinusitis (CRS) that can lead to pronounced morbidity. It has unique clinical and pathophysiological distinctions which make it different than other forms of CRS.^{1–3} Initial symptomatology is often characterized by seemingly innocuous nasal congestion, nasal obstruction, postnasal drainage, and anosmia.^{1–3} As the disease advances, bony erosion and extension of fungal debris into the intracranial cavity and orbit can ensue.^{4–6} At this point, patients present with more serious findings, such as visual disturbances, proptosis, and mental status changes. Patients with AFRS are typically young immunocompetent adults.³

Characteristic radiologic findings are well established, including total or near total opacification of the sinuses.

Dense heterogenous opacifications with areas of hyperattenuation are very commonly seen on CT scans.⁷ Bony erosion can also be seen in 20 to 56% of patients with AFRS.⁶ Recent study⁸ also showed that sinonasal variants, like concha bullosa, can be more prevalent in

¹Department of Otolaryngology, West Virginia University, Morgantown, West Virginia

²Department of Otolaryngology, Medical College of Georgia, Augusta University, Augusta, Georgia

³Department of Otolaryngology, Drexel University, Philadelphia, Pennsylvania

Corresponding Author:

Chadi A. Makary, Department of Otolaryngology, West Virginia University, Morgantown, WV 26506, USA..

Email: cmakary@hsc.wvu.edu

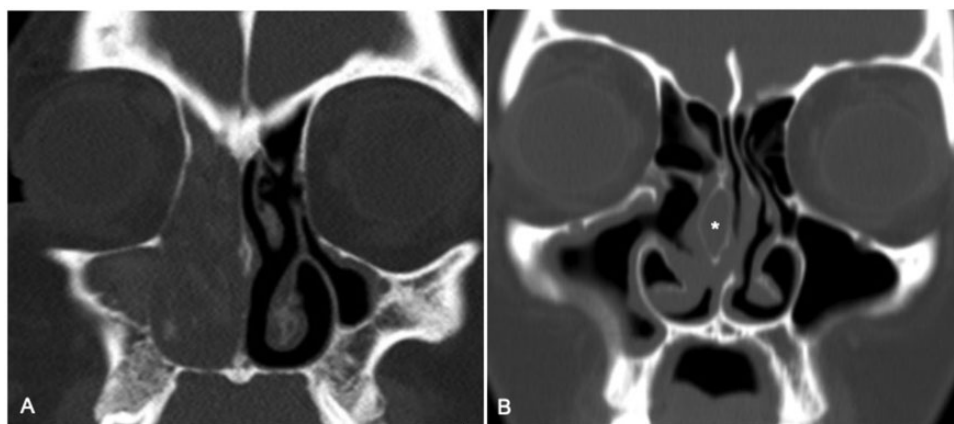


Figure 1. CT scans, bone window, coronal cuts of two patients with unilateral AFRS. A, Extensive disease which doesn't allow determination of any AV. B, Right sided opacified CB.

AFRS compared to other type of chronic rhinosinusitis (CRS) which according to them could explain the higher recurrence rate and need for revision endoscopic sinus surgery (ESS) in AFRS compared to other types of CRS.⁸

In this study, we aim to look at the prevalence of anterior sinonasal anatomic variants (AVs) in AFRS patients. Our goal is look at the prevalence of concha bullosa (CB), Haller cells (HC), Agger Nasi cells (AN), and supraorbital ethmoid cells (SOEC) in patients with unilateral AFRS. The healthy side in these patients will be used as their own control.

Materials and Methods

The Augusta University Institutional Review Board granted approval for this study. A prospectively collected database from the senior author's (SEK) academic tertiary-referral rhinology practice was searched and analyzed for all patients who presented with AFRS from 2008–2019. Patients with AFRS met diagnostic criteria as defined by the International Consensus Statement on Allergy and Rhinology: Rhinosinusitis,⁹ with type I hypersensitivity determined by formal testing or a reported history of environmental allergies. Those patients were reviewed for disease laterality. All patients with unilateral AFRS were included, and their preoperative CT scan of the sinuses were reviewed for the presence of anterior sinonasal anatomic variants (AVs). The scans reviewed were those prior to any ESS done (prior to first ESS in patients who had more than one surgery). AVs included concha bullosa (CB), infraorbital cells a.k.a. a Haller's cells (HC), Agger Nasi cells (AN), and supraorbital ethmoid cells (SOEC). All CT scans were obtained with 1 mm fine cuts slices (Figure 1). Scans were reviewed in axial, coronal and sagittal planes by a single reviewer, the senior author (SEK), to ensure reliability. Each patient's uninvolved side served as its own

Table 1. Demographic Data.

	Right	Left	Total (%)
Age (years)	28.8	35.4	32.5
Sex			
Male	5	5	10 (33.3%)
Female	9	11	20 (66.7%)
Race			
Black	10	9	19 (63.3%)
White	4	7	11 (36.7%)

control. In some cases, the extent of disease precludes determination of AVs. Those patients were excluded.

Statistical analysis was performed using the IBM SPSS statistical software version 25. Descriptive statistics of patient demographics were performed, and chi-square test was used to evaluate demographics. For all variables reported as averages, the Student t-test analysis was used to determine statistical significance. A P value of <.05 was considered to indicate a statistically significant difference for all statistical tests.

Results

A total of 155 patients with AFRS were identified. 30 patients (19.4%) had unilateral AFRS with scans showing recognizable AVs. Of those, 16 patients had left sided and 14 patients had right sided disease. Demographic data on these 30 patients are summarized in Table 1. The mean age was 32.5 years (range of 12–74 years), there were 20 female patients (66.7%), and 19 patients (63.3%) were African Americans. AVs were present in 26 of the 30 AFRS sides (86.7%) and in only 21 of the 30 healthy sides (70%), (chi squared = 2.45, $p = 0.117$) (Table 2). CB were present in 12 of 30 AFRS sides (40%) and only in 1 of 30 healthy sides (3%),

Table 2. All Anatomic Variants.

AFRS (N)	AFRS Side	Normal Side	P Value
Right (14)	12 (85.7%)	9 (64.3%)	0.117
Left (16)	14 (87.5%)	12 (75%)	
Total (30)	26 (86.7%)	21 (70%)	

(chi squared = 11.88, $p = 0.0006$) (Table 3). The presence of HC was more common in the AFRS vs healthy sides (7 of 30, 23.3% vs 2 of 30, 6.7%, respectively, $p = 0.071$) (Table 4). AN and SOEC presence was similar in both AFRS and healthy sides ($p = 0.598$ and $p = 0.718$ respectively) (Tables 5 and 6).

Discussion

AFRS is a distinct type of CRS, characterized by a set of unique radiographic and pathophysiological findings.¹⁻³ It occurs in immunocompetent individual, but the exact pathophysiology is not fully elucidated. AFRS occurs unilaterally in up to 20% of cases, certainly more than other types of CRS, including CRS with nasal polyps (CRSwNP). In our series, 19.4% of patients had unilateral disease. It is unknown why AFRS tends to occur unilaterally.

The tendency for AFRS to recur is high, requiring revision ESS, with the incidence reported to be up to 50% in some studies.^{10,11} It is imperative to address and completely open the CBs during surgery for AFRS as it can harbor leftover fungal debris.

Our study shows that sinonasal AV are more common in the sides with AFRS compared to the healthy sides of patients with unilateral disease. The importance of our study is that each patient served as its own control. CB is more prevalent in AFRS compared to other types of CRS, but it also more common on the side of disease in cases of unilateral AFRS. This study also shows that Haller cells are more common on the AFRS side compared to healthy sides, although it didn't reach statistical significance. The combination of both CB and HC may provide nidus for the fungal debris, which when combined with the obstructive effects of these AVs, can lead to the development of AFRS.

Prior studies failed to provide any association between the obstructive effects of CB and CRS development.¹²⁻¹⁵ In fact, the prevalence of CB in general is reported anywhere from 20 to 50%.^{16,17} However, the hypothesis is that AFRS is both a distinct phenotype and potentially endotype or endotypes under a broader group of CRSwNP.¹⁸ Therefore, what is true for CRS with and without nasal polyps can't be applied to AFRS. More studies are needed to determine the obstructive effects of AVs on AFRS development.

Table 3. Concha Bullosa.

AFRS (N)	AFRS Side	Normal Side	P Value
Right (14)	5 (35.7%)	0	0.0006
Left (16)	7 (43.8%)	1 (6.3%)	
Total (30)	12 (40%)	1 (3.3%)	

Table 4. Haller Cells.

AFRS (N)	AFRS Side	Normal Side	P Value
Right (14)	4 (28.5%)	1 (7.2%)	0.071
Left (16)	3 (18.6%)	1 (6.3%)	
Total (30)	7 (23.3%)	2 (6.7%)	

Table 5. Agger Nasi Cells.

AFRS (N)	AFRS Side	NORMAL SIDE	P Value
Right (14)	7 (50%)	8 (57.1%)	0.598
Left (16)	12 (75%)	9 (56.3%)	
Total (30)	19 (63.3%)	17 (56.6%)	

Table 6. Supraorbital Ethmoid Cells.

AFRS (N)	AFRS Side	Normal Side	P Value
Right (14)	1 (7.1%)	1 (7.1%)	0.718
Left (16)	4 (25%)	3 (18.8%)	
Total (30)	5 (16.7%)	4 (13.3%)	

Our study has important limitations that deserve discussion. Our analysis was not based on a randomized, blinded controlled trial. It is a retrospective review of data that was collected prospectively, which put it at risk for selection and information bias. A second limitation of the study is the small sample size of patients with unilateral AFRS. As this was a retrospective review of data, our cohort was limited by the number of patients in the database.

Conclusion

Anterior AVs are more common on the sides with AFRS compared to healthy sides, with the presence of CB reaching unquestionable statistical significance. Further studies are needed to determine the possible association of AVs, especially concha bullosa, with the pathophysiology of AFRS, and their effects on the surgical outcome and recurrence.

Authors' Note

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Declaration of Conflicting Interests

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ORCID iDs

Chadi A. Makary  <https://orcid.org/0000-0001-9967-4045>

Aykut Unsal  <https://orcid.org/0000-0002-6922-5561>

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