

Surgical Treatment of Benign Sinonasal Vascular Lesions: Hacettepe ENT Experience

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Abstract

Objective: Benign vascular lesions are common in the head and neck area, but sinonasal involvement is rare. This study aimed to present the features and surgical outcomes of patients with benign sinonasal vascular lesions in a single institution.

Material and Methods: Medical records of 20 patients who underwent surgery for benign sinonasal vascular lesions at the Department of Otolaryngology, Hacettepe University between 2005 and 2020, were retrospectively reviewed. Patients' demographic characteristics, lesion size and localization, surgical outcomes, and recurrence status were noted.

Results: The study group comprised 8 male (40%) and 12 female (60%) patients. Mean age was 41.3±22 years. The main presenting symptoms were epistaxis (60%) and nasal obstruction (30%). The most common sites of origin were the inferior turbinate (35%) and nasal septum (30%), followed by the maxillary sinus (15%). Mean lesion size was 27.1±13.2 mm. Histopathologically, 12 specimens were classified as vascular malformations and 8 as vascular tumors. Mean age was higher among the vascular malformations compared to the vascular tumors (49.9 y vs 28.5 y, p=0.031). All the lesions at the inferior turbinate were vascular malformations (p=0.014). The endoscopic endonasal technique was used in 16 patients (80%) whereas, combined techniques were used in 4 patients (20%). Residue/recurrence rate was 25%. The mean lesion size was larger in patients with residue/recurrence (42 mm vs 22 mm, p=0.001).

Conclusion: The endonasal endoscopic technique could be successfully used for the treatment of benign sinonasal vascular lesions. Lesion size was important with regard to residue/recurrence; thus, patients with larger lesions should be followed-up closely in the postoperative period.

Keywords: Hemangioma, vascular malformation, epistaxis, endoscopic sinus surgery, nasal

INTRODUCTION

The classification and denomination of benign vascular lesions in the literature have been confusing throughout the years. Inconsistencies in the terminology used between studies make it difficult to achieve a standardized clinical approach for the management of these lesions. Historically, benign vascular lesions were classified according to the dominant vessel size (small diameter vessels as capillary; large diameter vessels as cavernous) (1). However, with the update of the International Society for the Study of Vascular Anomalies, vascular anomalies have been grouped into vascular tumors and vascular malformations (2). The so-called "cavernous hemangioma" is now categorized as a vascular malformation (2). Vascular tumors, which consist of benign, borderline, and malignant tumors, have distinct clinical, radiological, and pathological features than vascular malformations. Hemangioma is a true vascular tumor, which arises from the vascular endothelial cells and exhibits endothelial hyperplasia; however, vascular malformations are localized defects of vascular morphogenesis caused by abnormalities during vasculogenesis (3, 4). Infantile hemangioma and congenital hemangioma form the two subgroups of hemangiomas. Pyogenic granuloma, also known as lobular capillary hemangioma, is a relatively common vascular tumor. Although it was thought to be a reactive hyperplasia, recent studies have suggested that it is a real benign neoplastic process (5).

Although benign vascular lesions are rare in the sinonasal area, making the accurate diagnosis is important to avoid massive epistaxis due to unnecessary biopsy or from the lesion itself. The main presenting symptoms are nasal obstruction and bleeding (6). Treatment options include watchful waiting, intralesional injections, and surgery. Complete surgical excision is the best treatment method (6). Endoscopic, external, or combined techniques could be

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used, depending on the features of the lesion, the patient, and the surgeon's preference. This study aimed to present the clinical features and analyze the surgical outcomes of patients with benign sinonasal vascular lesions who were surgically treated in a single center.

MATERIAL AND METHODS

We retrospectively reviewed the medical records of 20 patients who underwent surgery for benign sinonasal vascular lesions at the Department of Otolaryngology, Hacettepe University School of Medicine between 2005 and 2020. Patients' demographic characteristics and symptoms, lesion size and localization, and surgical technique and outcomes were noted. The clinical data also included preoperative imaging and embolization status, postoperative residue/recurrence rates, and complications. This study was approved by the Hacettepe University Ethics Committee (Approval Date: March 01, 2016; Approval Number: GO 16/72-24).

All the patients underwent routine otolaryngologic examination, including fiberoptic nasopharyngoscopy. Preoperative computed tomography and magnetic resonance imaging scans were carried out for all the patients. Preoperative biopsy was avoided due to the bleeding risk, and the diagnosis was based on preoperative imaging and examination. All the patients underwent surgical treatment in our clinic, of whom 6 had previously been treated with silver nitrate cauterization in other centers and were referred to us due to unresolved symptoms.

Statistical Analysis

Statistical analysis was performed using IBM Statistical Package for the Social Sciences software version 23.0 for Windows (IBM SPSS Corp.; Armonk, NY, USA) and Microsoft Excel spreadsheet software (Microsoft; Redmond, Washington, USA). Descriptive analysis was performed, and categorical variables were compared using Pearson's chi square test and Fisher's exact test for small-sample data ($n < 5$). Normal distribution was tested using the Shapiro-Wilk normality test and normal distribution

parameters. Comparisons of the means between two groups were done using the independent T test. Non-parametric independent variables were compared using the Mann-Whitney U test. The level of statistical significance was set at $p < 0.05$. All reported p-values are two-sided.

RESULTS

Of the 20 patients, 8 were males (40%) and 12 were females (60%). Mean age was 41.3 ± 22 years (range, 4 months-75 years). The most common presenting symptoms were epistaxis (60%), nasal obstruction (30%), facial swelling (20%), and pain and swelling in the eye (10%). In one patient, the symptoms were associated with pregnancy.

Mean lesion size was 27.1 ± 13.2 mm (range, 5-50 mm). The lesion originated from the inferior turbinate in 7 patients (35%), nasal septum in 6 patients (30%), maxillary sinus in 3 patients (15%), ethmoid sinus in 2 patients (10%), sphenoid sinus in 1 patient (5%), and nasal bone in 1 patient (5%) (Figure 1).

Specimens were classified as vascular tumors ($n=8$) and vascular malformations ($n=12$). Vascular tumors consisted of infantile hemangioma ($n=1$), spindle-cell hemangioma ($n=1$), pyogenic granuloma ($n=5$), and Masson's tumor (Intravascular papillary endothelial hyperplasia) ($n=1$). Table 1 shows the comparison of patients according to the histological type.

Two patients underwent preoperative embolization: Patient 1 was a 21-year-old female with a large 46-mm tumor in the maxilla and patient 2 was a 4-month-old baby, who had a 41-mm tumor in the ethmoid cells extending to the orbital cavity. The endonasal endoscopic technique was used in 16 (80%) patients. Combined procedures were used in 4 patients due to the localization and size of the tumor (2 Caldwell-Luc, 1 Maxillectomy, and 1 Osteoplastic approach).

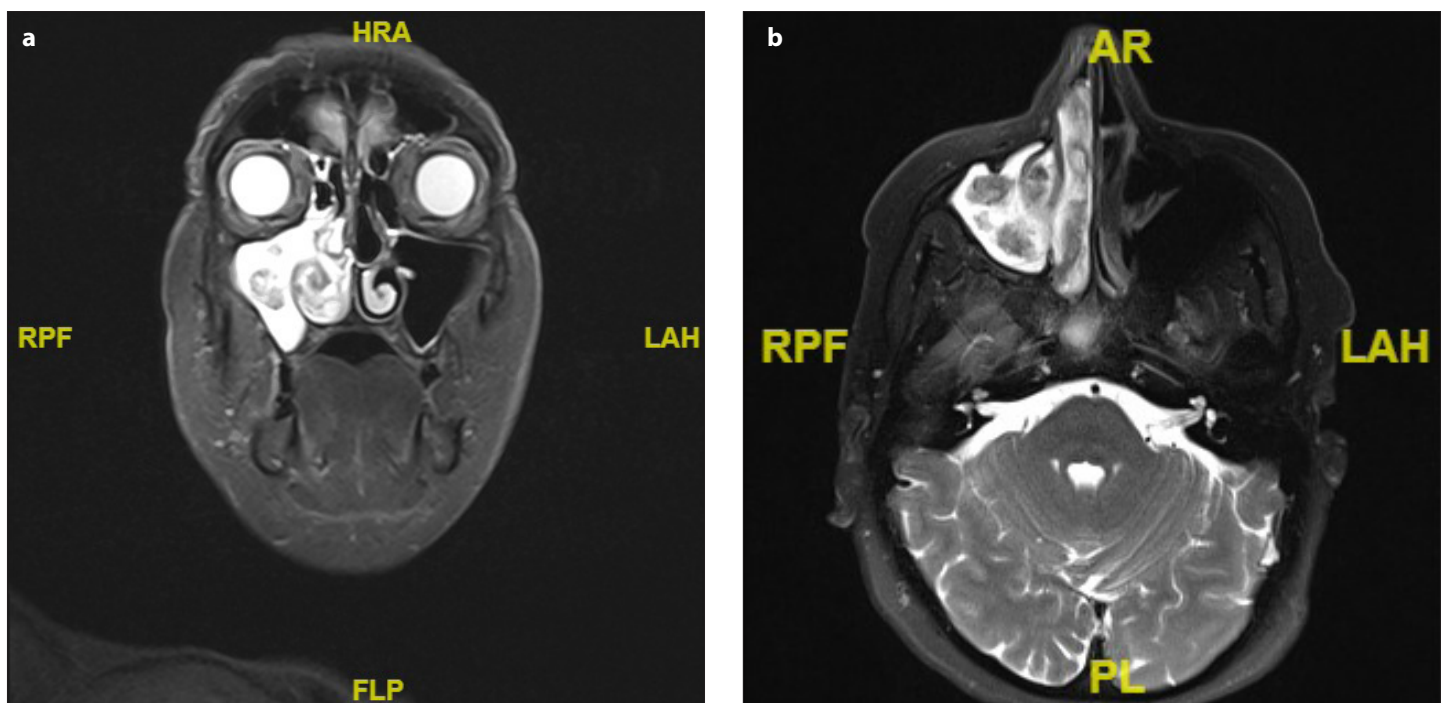


Figure 1. a, b. Coronal (a) and axial (b) sections of a 56-year-old female patient. A 4-cm mass originates from the inferior turbinate and extends laterally to the maxillary sinus

Table 1. Patient characteristics according to the histologic type

	All patients (n)	Vascular tumor	Vascular malformation	p
Age, years	41.3±22	28.5±24.6	49.9±16.3	0.031*
Gender, n				0.64
Male	8	4	4	
Female	12	4	8	
Lesion Size, mm (mean, standard deviation)	27.1±13.2	20.6±15	31.5±10.3	0.07
Follow-up, months median (range)	6 (1-132)	5 (1-132)	7 (1-132)	0.87
Lesion Localization, n				
Inferior turbinate	7	-	7	0.014*
Nasal septum	6	4	2	0.16
Maxillary sinus	3	2	1	0.53
Ethmoid sinus	2	1	1	1
Sphenoid sinus	1	-	1	1
Nasal bone	1	1	-	0.4
Surgical technique, n				0.25
Endoscopic	16	5	11	
Combined	4	3	1	

*Statistically significant.
n: number; mm: millimeters.

Median follow-up time was 6 months (range, 1-132 months). Complete resection could be achieved in 15 patients (75%). During the follow-up, 5 patients had postoperative residual/recurrent lesion and 4 of them underwent multiple surgeries due to unresolved symptoms. The mean time to reoperation was 3.7 months in the recurrent cases. The mean lesion size was significantly larger in patients with residue/recurrence (42 mm vs 22 mm, $p=0.001$). Two patients were diagnosed and treated at the pediatric age (<18 years old). Of these pediatric patients, one was a 4-month-old patient with spindle-cell hemangioma and the other one was a 7-month-old patient with infantile hemangioma. Residue/recurrence rate was not significantly different between pediatric and adult patients ($p=0.44$). Residue/recurrence rate was the highest (100%) in ethmoid localized lesions ($p=0.052$) (Figure 2). There were no intraoperative or postoperative complications. There was no significant difference between vascular tumors and malformations in terms of residue/recurrence ($p=1$). Table 2 summarizes the patients' features according to the residue/recurrence status and Table 3 shows the patients' features in terms of age groups.

DISCUSSION

Herein, we report our 15 years of experience in the surgical treatment and management of patients with benign sinonasal vascular lesions. The mean age of the patients in our group study (41.3 years) was in line with those of the previous reports (39 years and 40 years) (7, 8). Our patient group consisted of 12 females and 8 males (male to female ratio 1:1.5). However, there was no gender preponderance in the previous studies (7, 9). We think that this female preponderance is due to the small sample size used in our study. The main presenting symptoms were epistaxis and nasal obstruction (9, 10). The most common symptoms in our study

Table 2. Patient characteristics according to the recurrence status

	Residue/ Recurrent (n)	No residue/ Non-recurrent (n)	p
Age, years	38.2±29.8	42.4±20.3	0.72
Gender, n			1.0
Male	2	6	
Female	3	9	
Lesion size, mm (mean, standard deviation)	42±7.5	22.2±10.7	0.001*
Follow-up, months median (range)	12 (3-132)	6 (1-132)	0.45
Lesion localization, n			
Inferior turbinate	1	6	0.61
Nasal septum	1	5	1.0
Maxillary sinus	1	2	1.0
Ethmoid sinus	2	-	0.052
Sphenoid sinus	-	1	1.0
Nasal bone	-	1	1.0
Surgical technique, n			0.24
Endoscopic	3	13	
Combined	2	2	

*Statistically significant.
mm: millimeters; n: number.

cohort were similar with those of the previous studies (epistaxis 60% and nasal obstruction 30%).

Abnormal vascular endothelial proliferation is associated with the pathogenesis of vascular tumors, but their exact etiology is still not clear (11). Hormonal effects, trauma, viral oncogenes, and increase in growth factors are thought to play a role in the pathogenesis of these lesions (8). It is well known that pregnancy can promote the growth of vascular lesions or aggravate its symptoms. The incidence of vascular lesions during pregnancy is 2%-5% and it usually decreases after delivery (12). Nasal obstruction is a common symptom in pregnancy and can mask the clinical symptoms of an underlying vascular lesion. Thus, a thorough otolaryngologic examination should be performed and accompanying epistaxis should be questioned in the anamnesis. The decision to perform surgery during pregnancy is debatable, and should be undertaken in a patient-specific manner. Puxeddu et al. (8) reported two cases of pregnancy-associated sinonasal vascular lesions, who were operated under general anesthesia without any complication. Takaishi et al. (10) reported the case of a patient who had been previously operated for sinonasal benign vascular lesion and had a recurrence during her second pregnancy. We performed surgery on a pregnant woman who presented to our clinic with severe epistaxis and nasal obstruction, which had begun from the start of the pregnancy. Her symptoms had gradually become worse prior to her presentation at our clinic. Due to her 21-week-old pregnancy, we decided to operate her under local anesthesia and the lesion was endoscopically excised without any complication.

Our median follow-up time was 6 months (mean 28, range 1-132). The main outcome measure was our residue/recurrence rate which was 25%

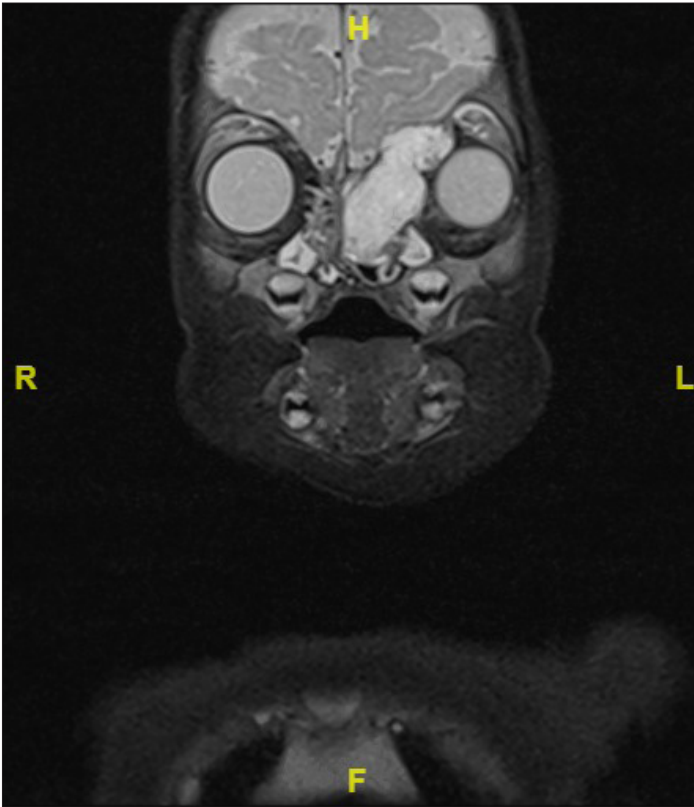


Figure 2. A 41-mm spindle-cell hemangioma in the ethmoid cells extending to the orbital cavity in a 4-month-old patient. He underwent a revision surgery 1 month after the initial surgery due to residue/recurrence

(5 patients). In contrast to some of the previous reports of Takaishi et al. (10) (3%) and Kim et al. (9) (5.4%), our recurrence was significantly higher. However, Smith et al. (7) reported 42% of recurrence in their lobular capillary hemangioma cohort. This inconsistency could be due to the heterogeneity of the patient groups and the relatively small sample size of our cohort. Our patients underwent reoperation at a mean of 3.7 months after the first operation; therefore, we think that the recurrences are probably due to the incomplete resection in the first operation. Larger tumor size could be a reason for the incomplete removal of the whole tumor. This was also supported by the difference in mean lesion size between the two groups (recurrent 42 ± 7.53 mm vs non-recurrent 22 ± 10.7 mm, $p=0.001$). Smith et al. (7) reported a significant recurrence rate among older patients; however, we could not show any difference in terms of age. Vascular malformations typically occur at the inferior turbinate or lateral nasal wall (9, 10). In line with this, all the lesions at the inferior turbinate in our cohort were vascular malformations, which reached statistical significance. Additionally, the mean age of the patients who had vascular tumors was lower than that of the patients with vascular malformations, probably because the 2 patients who had true hemangiomas were less than 1 year old.

Non-surgical treatment options for sinonasal benign vascular lesions consist of radiotherapy, electrocoagulation, embolization, silver nitrate, carbon dioxide snow, pulse-dye lasers, intra-lesional injections of sclerosing agents (sodium tetradecyl sulfate, sodium morrhuate, and sodium psylliate) (13). Recently, intralesional anti-VEGF (Bevacizumab) injection was also successfully used in a patient with recurrent sinonasal hemangioma (14).

One limitation of our study is the relatively small sample size. Another limitation is the retrospective design; however, given the rare nature of this lesion, it is hard to conduct prospective or large sample-sized studies in a single center. Further studies with larger sample sizes and standardized classifications are required to yield more statistically powerful results.

CONCLUSION

Benign vascular masses are relatively uncommon in the sinonasal area. Correct diagnosis and treatment is crucial to avoid mismanagement of the patients. Surgery is the mainstay of the treatment options and the endonasal endoscopic technique could be successfully used in most of the patients. Larger lesions tend to recur frequently and should be followed-up closely in the postoperative period.

Ethics Committee Approval: Ethics committee approval was received for this study from the Hacettepe University Ethics Committee (Approval Date: March 01, 2016; Approval Number: GO 16/72-24).

Informed Consent: Informed consent was not obtained due to the nature of the study.

Peer-review: Externally peer-reviewed.

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Table 3. Distribution of patient features according to age groups

	Age≤18 y	18 y<Age<60 y	60 y≤Age
N. of patients	2	13	5
Gender, n			
Male	2	5	1
Female	-	8	4
Lesion size, mm	30±11	25±12	31.6±14
Follow-up, months median (range)	3 (3)	8 (3-96)	6 (1-132)
Lesion localization, n			
Inferior turbinate	-	6	1
Nasal septum	1	2	3
Maxillary sinus	-	-	3
Ethmoid sinus	1	-	1
Sphenoid sinus	-	1	-
Nasal bone	-	1	-
Surgical technique, n			
Endoscopic	2	10	4
Combined	-	3	1

y: years; n: number.

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