

## Cavernous Sinus Hemangioma: Diagnosis and Treatment

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### Abstract

Cavernous hemangiomas of the maxillary sinus are rare. Here, we describe the management of a rare case of this vascular tumor of the maxillary sinus. We propose that an effective treatment can be achieved by performing endoscopic sinus surgery, preceded by a pre-operative embolization.

**Keywords:** Maxillary sinus; Hemangioma; Embolization

**Abbreviations:** IT: Inferior Turbinate; NS: Nasal Septum; MW: Medial Wall; NT: Necrotic Tissue; PW: Posterior Wall; NLD: Nasolacrimal Duct

### Introduction

Cavernous hemangiomas are common vascular tumors of the head and neck region, although those arising in the nose [1] and particularly in the maxillary sinus [2-4] are exceedingly rare. To the best of our knowledge, less than 50 cases of sinonasal cavernous hemangiomas have been reported in adults since 1959 [4-7]. Hemangiomas are divided into capillary and cavernous types, depending on the size of the dominant vessel affected, as seen by microscopic examination. Capillary hemangiomas are more common and are composed of capillary-sized vessels, lined with flattened epithelium [5]; they usually arise from the nasal septum. On the other hand, cavernous hemangiomas are composed of large, endothelium-lined vascular spaces, and more likely occur on the lateral wall of the nasal cavity [7,8]. Here, we describe the management of a 33-year-old woman with a large cavernous hemangioma in the maxillary sinus, which we resected with endoscopic medial maxillectomy, after performing arterial embolization.

### Case Report

A 33-year-old woman presented with a one-year history of right-side nasal obstruction and rhinorrhoea. Otherwise, she did not have any other remarkable medical history, and she was a non-smoker. The patient complained of the recent onset of a sense of right cheek fullness; she did not mention any bleeding. She had previously been diagnosed with sinusitis and treated with antibiotic therapy with only partial improvement of her symptoms.

Anterior rhinoscopy of the right nasal cavity revealed a bulge arising from the right nasal wall and impinging on the septum. Endoscopic examination revealed a mass in her right nostril, arising from the maxillary sinus, occupying the middle meatus, and projecting to the nostril and choana. We then performed computed tomography (CT), contrast-enhanced magnetic resonance (MR) imaging, and arteriography of the facial skeleton. The CT scan revealed a mass in the right maxillary sinus, extending to the nasal cavity, and associated with bony remodelling in the medial sinus wall (Figures 1 and 2). MR scan showed a heterogeneous enhancement in gadolinium-enhanced T1-weighted images.

Considering the possibility of sinonasal malignancy, a biopsy was taken to obtain tissue of the mass from the nasal cavity. The biopsy showed cavernous hemangioma.

External carotid arteriography revealed tumor staining in the entire right maxillary sinus. Pre-embolization of the hemangioma indicated

that the main supply to the mass originated from the terminal branches of the right internal maxillary artery. The tumor, which was contrast-enhanced during the arterial phase, was a slow-flow vascular lesion, from which the contrast medium rapidly washed out into the right facial vein (Figure 3).

Twenty-four hours after the embolization treatment, we performed an endoscopic type II medial right maxillectomy. The tumor, arising from the right lateral wall, occupied the entire maxillary sinus and extended to the choana (Figure 4). The procedure, depicted in Figure 5, involved the removal of the medial wall of maxillary sinus, part of the inferior turbinate and careful exposure of the nasolacrimal duct to preserve its integrity (Figures 5A-5C). A diamond tipped burr was used. Once the peduncle had been identified in the posterior wall of the maxillary sinus, the tumor was removed through the oral cavity (Figures 5D-5G). Finally, we performed an en-block resection of the mucoperiosteal flap, covering the lateral, superior, and most of the inferior maxillary sinus wall, by drilling the region where the peduncle attached to the wall (Figures 5H and 5I), to prevent recurrence of the disease. No significant hemorrhage occurred during the surgery; we limited intraoperative bleeding to 200 ml. At the end of the procedure, a 45-degree endoscopic camera was used to evaluate the entire cavity. There was no evidence of residual lesion. A nasal pack was placed and removed three days after the procedure, and the patient was discharged.

Histopathological analysis of the resected tumor revealed cavernous hemangioma, which was 7 cm long (Figure 6). Microscopic examination showed dilated sinusoidal vascular channels in the stroma lining, accompanied with an interconnecting fibrous wall, which was covered with flat endothelium, as well as aggregation of vessel structures. The patient was followed with endoscopic examinations. At the three-month follow-up, there was no evidence of recurrence (Figure 7), and the patient remained asymptomatic. After about 14 months from surgery, the patient was yet asymptomatic and objectively free from disease. A new CT scan, at 14 months follow-up, showed sinus spaces free from disease (Figure 8).

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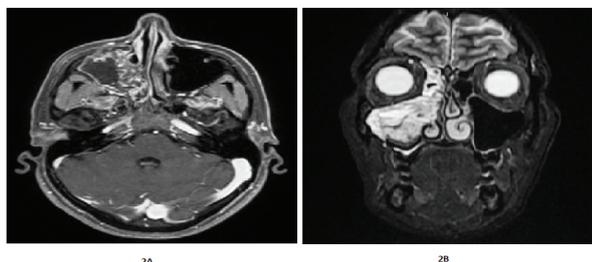
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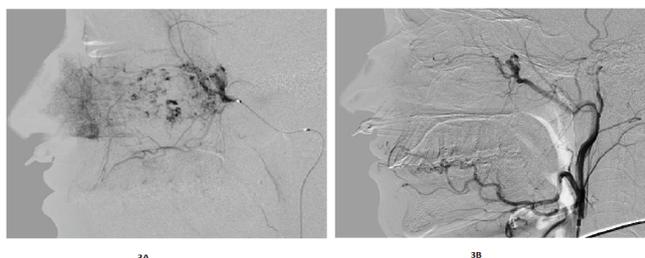
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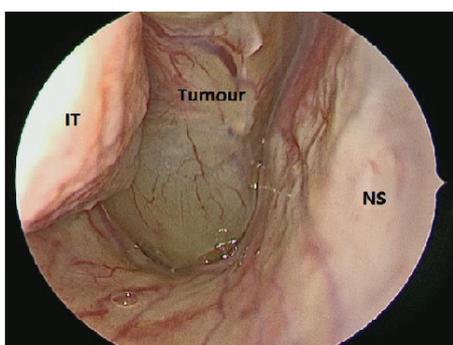
**Figure 1:** Pre-operative computed tomography scan (A) Axial and (B) Coronal view of the bone window showing a large soft-tissue mass occupying the right maxillary sinus and extending into the nasal cavity. The mass is associated with remodeling of the medial wall of the right maxillary sinus.



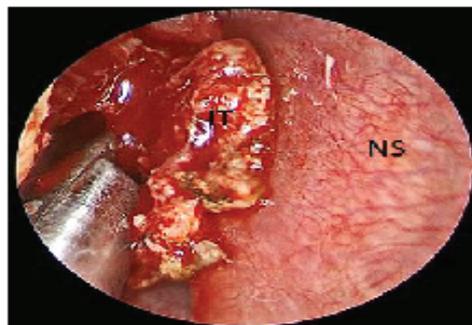
**Figure 2:** Pre-operative magnetic resonance imaging Gadolinium-enhanced T1-weighted magnetic resonance imaging (MRI) (A) and coronal short tau inversion recovery MRI (B) showing flow voids within a heterogeneously enhanced vascular tumor.



**Figure 3:** Pre- and post-embolization arteriography of the right internal maxillary artery. The pre-embolization image (A) shows the main supply to the mass, originating from the terminal branches of the right internal maxillary artery. The post-embolization image (B) shows a reduction of the tumor blood flow.

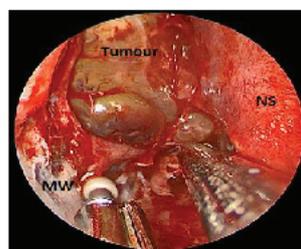


**Figure 4:** Pre-operative endoscopic view of the right nostril, endoscopic view at 0° with a 4-mm telescope.

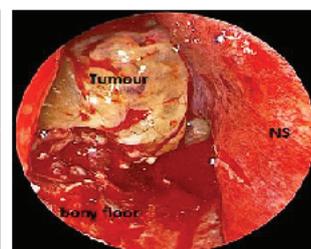


**5A**

**Figure 5A:** Step-wise endoscopic type II medial maxillectomy, endoscopic view of the inferior turbinate section.



**5B**



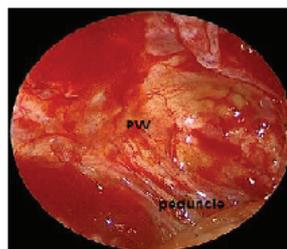
**5C**

**Figures 5B and 5C:** Endoscopic view of the maxillary sinus before (B) and after (C) drilling the medial wall.

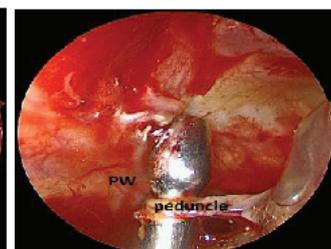


**5D**

**Figure 5D:** Endoscopic view of the relationship between tumor and mycosis.

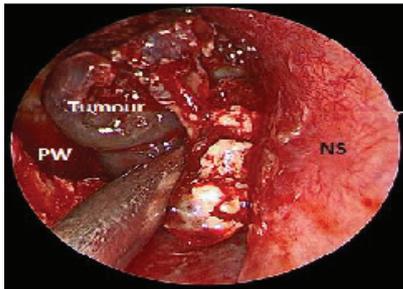


**5E**



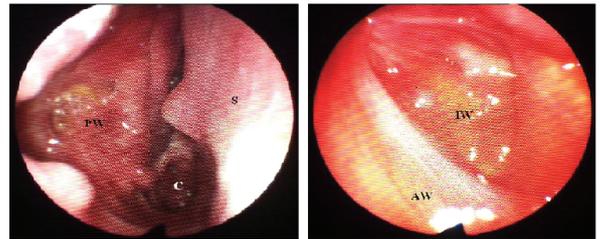
**5F**

**Figures 5E and 5F:** Endoscopic view of the attachment site of the peduncle in the posterior wall of the maxillary sinus (E) and its section (F).



5G

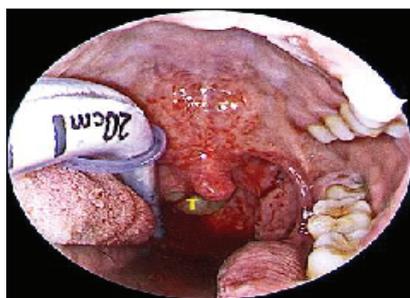
Figure 5G: View of the tumor after medial maxillectomy.



7A

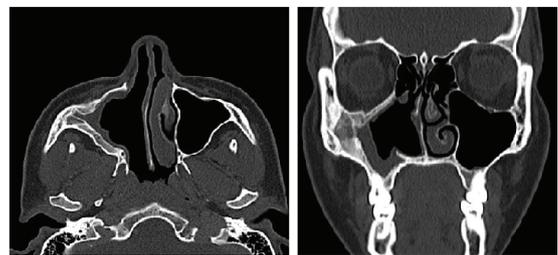
7B

Figure 7: Endoscopic view of the right nostril at an angle of 0°, three months after type II medial maxillectomy. (PW: Posterior Wall; S: Septum; AW: Anterior Wall; IW: Inferior wall).



5H

Figure 5H: View of the tumor through the oral cavity.



8A

8B

Figure 8: Post-operative computed tomography scan at 14 months follow-up (A) Axial and (B) Coronal view of the bone window showing the right maxillary sinus free from disease with only a mucosal thickening.



5I

Figure 5I: Endoscopic view of the maxillary sinus and the exposed nasolacrimal duct from a 45° angle.



Figure 6: Tumor after resection image showing that the resected tumor was 7 cm long.

## Discussion

Hemangiomas are common benign vascular tumors of the head and neck region commonly found in skin and mucosa but rarely found in the sinonasal soft tissue [3-9]. They are divided into capillary and cavernous types, depending on the size of the primary vessel affected. Capillary hemangiomas, usually arising in the nasal septum [10,11], are composed of capillary-sized vessels, which are lined with flattened epithelium, and are common, especially at a younger age.

Cavernous hemangiomas, on the other hand, are typically found in adults, and are characterized by larger, endothelium-lined vessels; they often occur on the lateral wall of the nasal cavity [12]. Hemangiomas are more prevalent in females, with a 5-6:1 Female-to-male ratio, whereas other vascular malformations occur independently of gender or race [13-15].

The exact origin of hemangiomas remains a topic of controversy. Some researchers have proposed the involvement of embryonic unipotent angioblastic cells, which could elongate and form blood vessels [16]. Although there have been multiple clinical studies of nasal hemangiomas [17], only 24 cases of maxillary hemangiomas have been reported [5,6]. Common symptoms of sinonasal hemangiomas are nasal obstruction, epistaxis, and, occasionally, a visible nasal mass [18]. In CT scans, cavernous hemangiomas appear as growing soft-tissue masses, expanding in air-containing spaces. Usually, they produce benign-appearing bone changes, followed by secondary expansion and thinning of bone structures [19]. According to Dillon et al., capillary hemangiomas have intense enhancement on MR imaging, and, in 50% of the cases, a peripheral hypo-intense rim, surrounding a central mixed mass, can be seen on T2-weighted images [19]. Consistently, our patient's MR scan also showed flow voids within a heterogeneously enhanced vascular tumor and a central necrotic hypointense area in T1-weighted images.

Differential diagnoses of sinonasal cavernous hemangiomas include inverted papilloma, mucocele, arteriovenous fistula, lymphangioma, glomangioma, angiofibroma, pyogenic granuloma, carcinoma, angiosarcoma, and hemangiopericytoma [20,21].

The traditional treatment for sinonasal hemangiomas is surgical excision with an open approach, in the form of craniofacial resection. However, recent advances in endoscopic surgical techniques have allowed for a less invasive removal of these tumors. Moreover, preoperative trans arterial embolization can decrease the tumor size and reduce the risk of hemorrhage during surgery [18]. In our patient, we performed a preoperative embolization of the right internal maxillary artery and subsequently a type II endoscopic medial maxillectomy, by which we effectively removed the tumor.

## Conclusion

In conclusion, we present the effective treatment of one of the few rare cases of maxillary hemangioma. We propose that endoscopic sinus surgery, along with the development of new techniques and preoperative embolization will greatly advance the treatment of this pathology.

## References

1. Fu YS, Perzin KH (1974) Non-epithelial tumors of the nasal cavity, paranasal sinuses and nasopharynx: A clinico-pathologic study of cartilaginous tumors (chondroma, chondrosarcoma). *Cancer* 34: 453-463.
2. Nishimura F, Young-Su P, Nakagawa I, Yamada S, Nakase H, et al. (2018) Hemorrhagic cavernous sinus hemangioma with sudden onset abducens palsy – A case report. *World Neurosurg* 8: 1878-8750.
3. Bakhsheshian J, Zada G, Pham MH (2018) Cavernous sinus hemangioma: A rare vascular tumor of cavernous sinus. *World Neurosurg* 110: 432-433.
4. Kim JS, Kwon SH (2017) Sinonasal hemangioma: Diagnosis, treatment and follow-up of 37 patients at a single centre. *J Oral Maxillofac Surg* 75: 1775-1783.
5. Takaishi S, Asaka D, Nakayama T, Iimura J, Matsuwaki Y, et al. (2017) Features of sinonasal hemangioma: A retrospective study of 31 cases. *Auris Nasus Larynx* 44: 719-723.
6. Sarkar S, Roychoudhury A, Roychoudhuri BK (2011) Intractable anemia: A case of bleeding nasal cavernous hemangioma. *Indian J Otolaryngol Head Neck Surg* 63: 23-24.
7. Osborn DA (1959) Haemangiomas of the nose. *J Laryngol Otol* 73: 174-179.
8. Weiss SW, Enzinger FM, Goldblum JR (2008) Enzinger and Weiss's soft tissue tumors. Mosby-Elsevier 303-324.
9. Sheppard LM, Mickelson SA (1990) Hemangiomas of the nasal septum and paranasal sinuses. *Henry Ford Hosp Med J* 38: 25-27.
10. Baki A (2018) Nasal septal cavernous hemangioma. *J Craniofac Surg* 29: 135-136.
11. Jammal H, Barakat F, Hadi U (2004) Maxillary sinus cavernous hemangioma: A rare entity. *Acta Otolaryngol* 124: 331-333.
12. Som PM, Brandwein MS (2003) Tumors and tumor like conditions: Head and neck imaging. Mosby 261-373.
13. Caylakli F, Cagici AC, Hurcan C, Bal N, Kizilkilic O, et al. (2008) Cavernous hemangioma of the middle turbinate: A case report. *Ear Nose Throat J* 87: 391-393.
14. Duvvuri U, Carrau RL, Kassam AB (2006) Vascular tumors of the head and neck. *Head and Neck Surgery-Otolaryngology*. Lippincott Williams & Wilkins, Philadelphia, PA, USA. pp: 1811-1825.
15. Mulliken JB, Glowacki J (1982) Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. *Plast Reconstr Surg* 69: 412-422.
16. Gupta DTK, Brasfield RD (1972) Blood and lymph vessel tumors. *CA Cancer J Clin* 22: 176-183.
17. Iwata N, Hattori K, Nakagawa T, Tsujimura T (2002) Hemangioma of the nasal cavity: A clinico-pathologic study. *Auris Nasus Larynx* 29: 335-339.
18. Song CE, Cho JH, Kim SY, Kim SW, Kim BG, et al. (2009) Endoscopic resection of haemangiomas in the sinonasal cavity. *J Laryngol Otol* 123: 868-872.
19. Dillon WP, Som PM, Rosenau W (1991) Hemangioma of the nasal vault: MR and CT features. *Radiol* 180: 761-765.
20. Dahodwala MQ, Husain Q, Kanumuri VV, Choudhry OJ, Liu JK, et al. (2013) Management of sinonasal hemangiopericytomas: a systematic review. *Int Forum Allergy Rhinol* 3: 581-587.
21. Tessema B, Eloy JA, Folbe AJ, Anstead AS, Mirani NM, et al. (2012) Endoscopic management of sinonasal hemangiopericytoma. *Otolaryngol Head Neck Surg* 146: 483-486.