Case Report

Mucosal Cavernous Hemangioma of the Maxillary Sinus

Mainak Dutta MS•[1], Sohag Kundu MS[1], Sabyasachi Barik MBBS[1], Shoham Banerjee MS[2], Subrata Mukhopadhyay MS[1]

Abstract

Mucosal cavernous hemangiomas of maxillary sinus and the lateral nasal wall are seldom encountered and difficult to diagnose with misleading radiologic features like bone erosion and heterogeneity due to patchy contrast uptake. The overall picture mimicking sinonasal malignancy, it is unclear whether there is true breach in the bone or remodeling due to the lesion's chronicity. Interestingly, it often does not bleed as expected during surgery, questioning the use of therapeutic embolization and pre-intervention vascular shrinkage. The clinical presentation and management protocol of sinonasal cavernous hemangiomas seem greatly individualized. We here present a patient with cavernous hemangioma of maxillary sinus and discuss the distinguishing clinical, histologic and imaging characteristics and subsequent management options, and attempt to establish the findings as the basis of considering it as an important differential diagnosis of radiologically heterogeneous sinonasal mass with suspected bone erosions presenting with nasal obstruction and epistaxis, mostly in young women.

Keywords: Cavernous hemangioma, epistaxis, hemangioma, maxillary sinus, paranasal sinus

Cite this article as: Dutta M, Kundu S, Barik S, Banerjee S, Mukhopadhyay S. Mucosal Cavernous Hemangioma of the Maxillary Sinus. *Arch Iran Med*. 2015; **18(2):** 130 – 132.

Introduction

Hemangiomas, the commonest vascular lesions of the head and neck, are slow-growing benign pseudotumoral vascular anomalies, or hamartomas, and are of capillary, cavernous, mixed and proliferative types. Although the face, scalp, orbit and oral cavity are mostly affected in the head-neck region, involvement of the nasal cavity and paranasal sinuses is very rare. The majority of the sinonasal hemangiomas are of capillary variety, usually seen in childhood in the septum and vestibule,[1,2] and the less common cavernous type,[3] generally encountered in adults with a female preponderance,[1,2,4–8] characteristically involves the lateral nasal wall.[4] Such hemangiomas can arise from the bone, mucosa or submucosa,[1] and very few cases of non-osseous (mucosal) variety of cavernous hemangioma originating from the maxillary sinus are reported in the indexed medical literature. Clinically and radiologically, they are difficult to diagnose, because due to the scarcity of cases a comprehensive clinical picture is lacking, and the imaging results are often non-specific, increasing the chances of misinterpretation with the more common sinonasal disorders.

Case Report

A 24-year-old woman presented with occasional nose-bleed since 18 months and gradually-progressive right-sided nasal obstruction for six months. The epistaxis was from her right nasal cavity, intermittent but profuse, and was not associated with any purulence, crusting, and nasal discharge. Anterior rhinoscopy of the right nasal cavity revealed a slough-covered, firm painless mass with few blood clots. On nasal endoscopy, the nasal cavity was found occupied by a large polypoid bluish-grey mass that bled on touch (Figure 1). High resolution plain and contrast-enhanced computed tomography (CECT) showed complete opacification of the right maxillary sinus with collections in ethmoid and frontal sinuses, widening of the ostiomeatal complex and destruction of the medial wall of maxilla (Figure 2a). The mass filled the entire nasal cavity up to the choana. Uneven uptake of contrast rendered the lesion inhomogeneous (Figure 2b).

Considering the possibility of sinonasal malignancy, a punch biopsy was planned. Routine hematological examinations, including coagulation profile, were unremarkable. An endoscopic biopsy from the nasal mass, however, showed dilated anastomosing blood vessels lined by endothelium, with no features of malignancy (Figure 3). The histologic picture suggested cavernous hemangioma, but interestingly there was no appreciable bleeding during or following the diagnostic procedure. The tumor was removed completely by combined approach endoscopic sinus surgery. Histopathology corroborated the biopsy findings. The patient recuperated well with no further bleeding in the immediate and late post-operative period, and was disease-free on two-years follow-up.

Discussion

Cavernous hemangiomas rarely involve the nose and paranasal sinuses, with only a handful of isolated case records that describe them especially in relation to the maxillary sinus and lateral nasal wall.[1,2,4–12] They are often asymptomatic,[9] or present as reddish, polypoid or sessile mass causing nasal obstruction or recurrent epistaxis[1] with a predilection for young persons, especially women, in their second and third decades.[4–8] Other presenting symptoms could be rhinorrhea, facial swelling and bulging of the eye.[1] Considering the scarcity of cases and dearth of narrative reviews, there is no generalized clinical picture or uniform management protocol for maxillary cavernous hemangiomas, thereby increasing the chance of misdiagnosis, and as a result, unpredictable bleeding during interventions.

Authors' affiliations: [1]Department of Otorhinolaryngology and Head-Neck Surgery, Medical College and Hospital, West Bengal, India. [2]Department of Otorhinolaryngology and Head-Neck Surgery, Midnapore Medical College and Hospital, Midnapore, West Bengal, India

• **Corresponding author and reprints:** Department of Otorhinolaryngology and Head-Neck Surgery, Medical College and Hospital, 88, College Street, Kolkata – 700073, West Bengal, India. Tel: +91-9830737587,
E-mail: duttamainak@yahoo.com
Accepted for publication: 9 November 2014

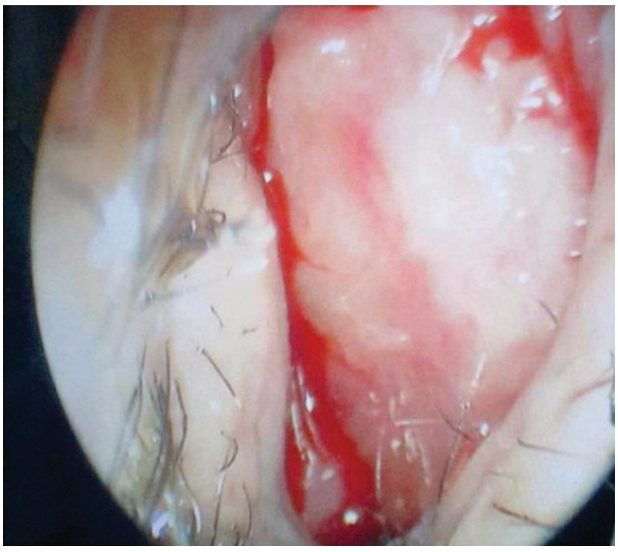

Figure 1. Endoscopic view of the hemangioma occluding the right nasal cavity. Note the bleeding around the lesion.

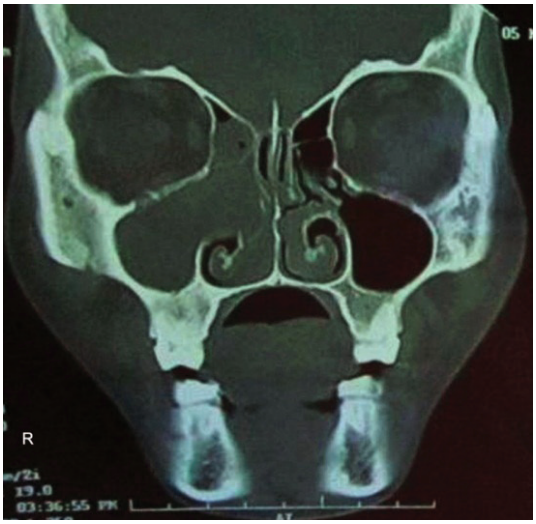

Figure 2a. Non-contrast CT-scan showing the mass occupying the right maxillary sinus and involving the ipsilateral nasal cavity with widening of the ostiomeatal complex.

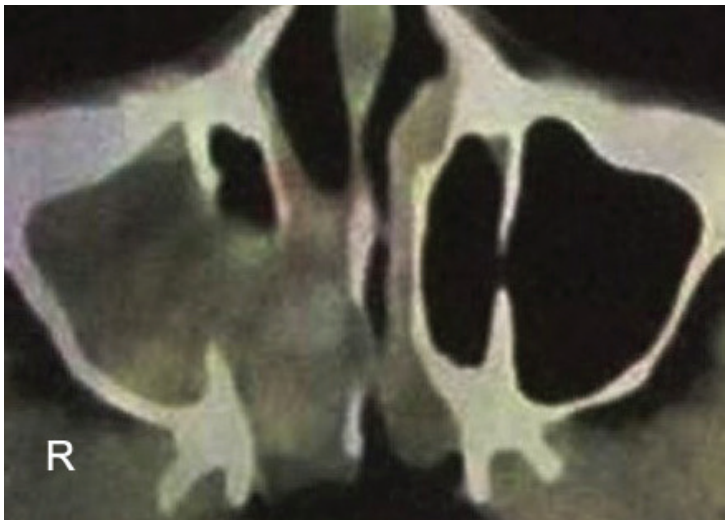

Figure 2b. CECT showing heterogeneous right-sided sinonasal mass extending up to the choana with patchy areas of contrast uptake.

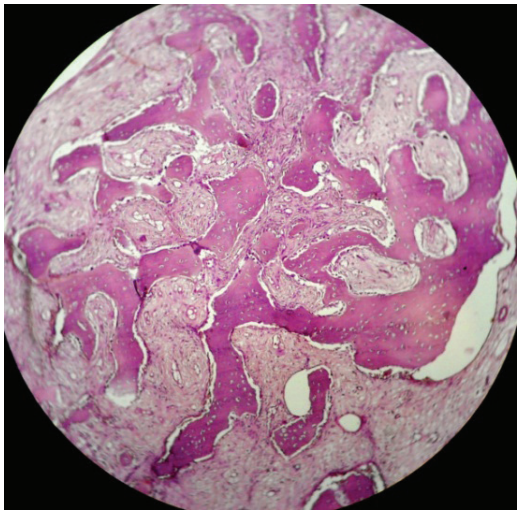

Figure 3. Photomicrograph of the lesion showing large, dilated vessels with sinusoidal blood-filled intercommunicating cavities arranged in lobular or diffuse pattern and lined by single layer of flattened endothelium [Hematoxylin-Eosin; 100×]

While the osseous variety remains confined within the bony trabeculae, the mucosal cavernous hemangioma presents as well-demarcated complete opacification of the sinus cavity in CT-scan with variable extensions. There are no pathognomonic radiologic findings, but as we have seen in our patient, there might be bulging of the medial wall of maxilla, widening of ostiomeatal complex (the area where it is said to originate), and involvement of adjacent sinuses like the ethmoids. The resultant bony deformity has been referred to as erosions in most records.[1,4,6] However, because cavernous hemangiomas are quiescent or slow-growing benign lesions, and can have compressive mass effect,[6] it is not clear whether they are true erosions or bone remodeling. True bone erosions, often significant, have been documented on rare occasions on involvement of the orbital floor, posterior wall of the maxillary sinus, and invasion within the ethmoids and sphenoid.[1,2] Nevertheless, a hemangioma is seldom suspected, and a large unilateral mass in the maxillary sinus communicating with the nasal cavity with epistaxis and apparent erosive features on imaging would most likely be considered malignant.[1,2,4,6] The dilemma is compounded when it takes up contrast in patches due to intralesional necrotic foci and extravasated clots that remain un-enhanced in the CT[2] and misinterpreted as necrosis in a malignant tumor. In magnetic resonance imaging, these areas appear hyperintense in T2-weighted images with an overall heterogeneous background.[1] The other differential diagnoses could be inverted papilloma,[1,2] pyogenic granuloma, mucoceles,[1,6] polypoid cystic masses,[1,2] and vascular tumors of the paranasal sinuses like angiosarcoma,[6] hemangiopericytoma[6] and Kaposi's sarcoma, and also neuroma when it remains proximal to the infra-orbital canal. Occasionally, it may be confused with invasive bony hemangiomas originating from nasal bones and maxilla. Correct pre-operative diagnosis is therefore of immense importance as each of these conditions has distinct clinical course and management guidelines.

Histologically, cavernous hemangiomas are composed of large, dilated vessels having sinusoidal blood-filled intercommunicating cavities arranged in lobular or diffuse pattern and lined by a single layer of flattened endothelium. The distinction between the cavernous and capillary types is not always well-defined as the microscopic size of the predominant vessels can vary in different areas of the tumor. In any case, cavernous hemangiomas are venous malformations supplied by small-to-medium-sized vessels with a low flow or non-arterial circulation. This might explain, like in our patient, why they occasionally do not bleed as expected during diagnostic and surgical procedures.[2] Angiography may provide an idea regarding vascularity as it results in pooling of contrast in the lesion[2,6] and persistence of vascular blush in late films, and also help prevent unpredictable bleeding during intervention with selective transarterial embolization.[2,6] It appears that its implementation, especially for therapeutic purpose, is individualized as there are reports where such lesions have been excised without prior vascular shrinkage and troublesome per-operative bleeding.[6]

Cavernous hemangiomas may regress spontaneously. In symptomatic patients, treatment is primarily surgical, ranging from complete excision to local resection sparing vital structures[6,9]; the approaches could be open, endoscopic or combined depending on the situation and extent of the lesion. Hemangiomas are generally radioresistant − steroid therapy and laser excision may be effective but have not yet enjoyed routine use.[9] Overall, the prognosis is encouraging. Though data from long-term follow-up is missing, it seems evident that cavernous hemangiomas seldom recur following complete excision, and malignant transformation is unknown. Yet a high index of suspicion is needed to consider a sinonasal mass as hemangioma, chiefly because its often-dramatic presentation, inconclusive preoperative diagnosis, rarity, and an impression of bone erosion on imaging closely simulate the clinical picture of sinonasal malignancy. Most importantly, the chances of bleeding during intervention remains unpredictable, with the indications of pre-operative catheter embolization still not clear. Mucosal cavernous hemangiomas of the maxillary sinus/lateral nasal wall are truly rare though well-described entities; understanding their clinico-pathologic profile would help identify these benign neoplasms as one the chief differential diagnoses of sinonasal neoplastic mass that presents mostly in young patients with nasal obstruction and epistaxis.

Conflict of interest: None declared.

Financial disclosure: Nil.

References

1. Vargas MC, Castillo M. Sinonasal cavernous haemangioma: a case report. *Dentomaxillofac Radiol*. 2012; **41:** 340 – 341.
2. Kim HJ, Kim JH, Kim JH, Hwang EG. Bone Erosion Caused by Sinonasal Cavernous Hemangioma: CT Findings in Two Patients. *AJNR Am J Neuroradiol*. 1995; **16:** 1176 – 1178.
3. Som PM, Brandwein MS. Tumors and tumor-like conditions. In: Som PM, Curtin HD, eds. *Head and Neck Imaging*. St. Louis, MO: Mosby; 2003: 261 – 373.
4. Hamdan AL, Kahwaji G, Mahfoud L, Husseini S. Cavernous hemagioma of the maxillary sinus: a rare cause of epistaxis. *Middle East J Anesthesiol*. 2012; **21(5):** 757 – 760.
5. Louza JP, Gürkov R, Weiler C, Krause E. Unilateral maxillary sinusitis: a cavernous haemangioma with bone destruction [in German]. *HNO*. 2011; **59(7):** 696 – 699.
6. Mussak E, Lin J, Prasad M. Cavernous hemangioma of the maxillary sinus with bone erosion. *Ear Nose Throat J*. 2007; **86(9):** 565 – 566.
7. Starska K, Lukomski M, Danilewicz M. Hemangioma cavernous of the nasal cavity and the maxillary sinus--case report and review of the literature [in Polish]. *Otolaryngol Pol*. 2006; **60(6):** 955 – 957.
8. Slaba S, Karam R, Hokayem N, Haddad S, Abadjian G, Aoun N, et al. Capillarovenous angioma of the maxillary sinus. Apropos of a case with review of the literature [in French]. *Ann Otolaryngol Chir Cervicofac*. 1997; **114(6):** 231 – 234.
9. Jammal H, Barakat F, Hadi U. Maxillary sinus cavernous hemangioma: a rare entity. *Acta Otolaryngol*. 2004; **124(3):** 331 – 333.
10. Engels T, Schörner W, Felix R, Witt H, Jahnke V. Cavernous hemangioma of the maxillary sinus [in German]. *HNO*. 1990; **38(9):** 342 – 344.
11. Amor Dorado JC, Juiz P, Zubizarreta A, Rossi J, Pulpeiro JR. Cavernous hemangioma of the maxillary sinus [in Spanish]. *Acta Otorrinolaringol Esp*. 1998; **49(2):** 165 – 167.
12. Lahoz Zamarro MT, Galvé Royo A, Royo J, Valero J, Cámara F. Cavernous hemangioma of the maxillary sinus [in Spanish]. *Acta Otorrinolaringol Esp*. 1990; **41(3):** 197 – 199.