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CAVERNOUS HEMANGIOMA OF MAXILLARY SINUS: CASE REPORT OF A RARE DISEASE AND LITERATURE REVIEW

ABSTRACT

The most common site for the nasal hemangioma to originate is nasal septum followed by the lateral nasal wall and vestibule. Cavernous hemangioma of the maxillary sinus is a rare presentation.This article reports a case of 65 years old lady with cavernous hemangioma of the left maxillary sinus with bone eroding properties in the imaging mimicking a malignant lesion.

Keywords: Bone erosion, Cavernous hemangioma, Maxillary sinus.

INTRODUCTION

Hemangioma is a benign neoplasm originating from the vascular tissue of skin, mucosa, bone, muscles and glands.[1] Hemangioma of the nasal cavity arises most commonly from the septum (65%), lateral wall (18%), and vestibule (16%).[2] Nasal hemangiomahas been classified as capillary, cavernous, mixed and proliferative types. The majority of sinonasalhemangiomas are capillary, whereas the cavernous type is extremely rare.The cavernous hemangiomas are common in adults and are more frequently seen in women.[3] It usually presents as unilateral epistaxis with nasal obstruction.

To our knowledge, cavernous hemangioma arising from the maxillary sinus is rarely reported in the literature. We report a case of cavernous hemangioma involving the left maxillary sinus treated by trans-nasal endoscopic excision technique.

CASE REPORT

A 65 years old female presented tothe ENT OPD with progressive left nasal obstruction for three years associated with intermittent unprovoked bleeding from the left nasal cavity for six months and protrusion of mass from the left nasal cavity for one day.

On examination, approximately 2x1 cm reddish-brown, globular mass with crust on its lower surface was protruding from the left nasal cavityalong with blood-tinged mucoid discharge (Figure I). There was fullness present in the mid and lower one-third of the left nasofacial groove with a broad nasal dorsum. Nasal airway patency was absent on the left side and inadequate on the right side. Anterior rhinoscopyon the left side revealed a 4x2 cm single, reddish-brown, globular mass covering the nasal cavity completely with blood-tinged mucoid nasal discharge. On the right side, there was a gross deviated nasal septum.

On probing, the mass was soft, mobile, sensitive and bled on touch.Theprobe could be passedinferiorly, medially and superiorly. The lateral passage was restricted.

On posterior rhinoscopy, a proliferative, irregular mass was seen covering both the choana with no visualisation of other normal structures.Sensation over the distribution of the Trigeminal nerve was preserved.

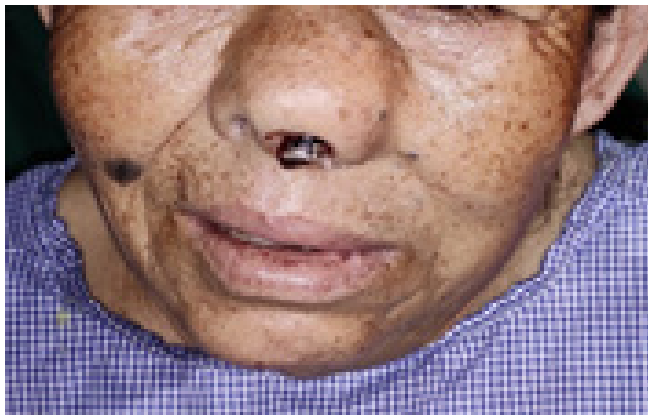

Figure I: Mass protruding from the left nasal cavity

Nasal endoscopy could not be done since the mass filled the entire nasal cavity up to the choana.She gave a history of biopsy taken from the left nasal cavity multiple times which was inconclusive. She was admitted to our hospital two times following

biopsy due to profuse bleeding and it was managed by nasal packing and bipolar cautery.

A high-resolution CECT scan showedheterogeneously enhancing soft tissue lesion in the left maxillary sinus extending into the nasal cavity and choana with an erosion of the nasal septum, medial wall of the maxilla,floor of the left maxillary sinus with the widening of the osteomeatal complex.Uneven uptake of contrast rendered the lesion inhomogeneous (Figure II).

Considering the possibility of sinonasal malignancy being a unilateral pathology, four times punch biopsy was taken which turned out to be inconclusive but negative for malignancy. Routine haematological examinations, including coagulation profile, were unremarkable.

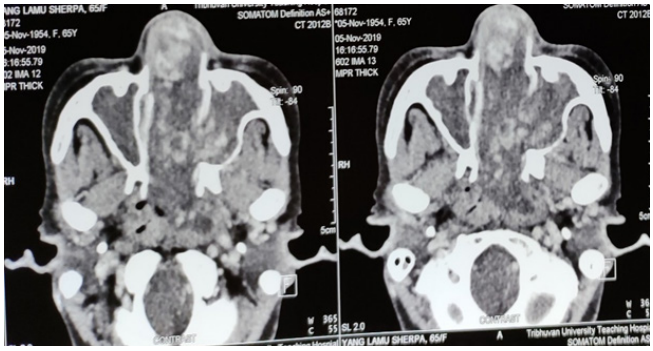

Figure II: Axial view on CECT scan showing a heterogeneously enhancing lesion in the left nasal cavity and maxillary sinus

We considered sinonasal malignancy or a tumour of vascular origin as the differential diagnosis. We did a preoperative angiography and embolized the feeding vessel which was the internal maxillary artery in this case. After preoperative work-up, under hypotensive anaesthesia, the lesion was excised viaa trans-nasal endoscopic approach. During surgery, a dark greyish and lobulated soft tissue was seen (Figure III) with its attachment to the posterior wall of the left maxillary antrum, left posterior fontanellaeand occupying left maxillary antrum, left nasal cavity, choana, nasal vestibule and protruding out of the left nostril. The mass was excised in a piecemeal manner and sent for histopathology. The intraoperative blood loss was roughly 100 ml.

The postoperative course was uneventful and the nasal pack was removed on the 2nd post-operative day. On following up with the patient, the histopathology reported of the excised specimen came out tobe consistent with cavernous

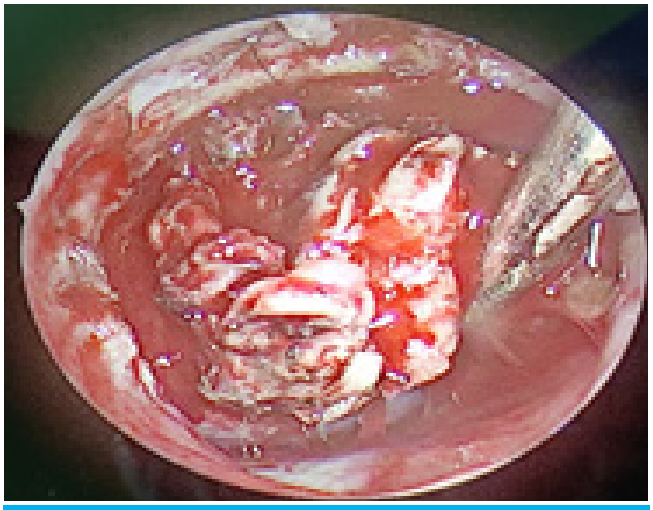

Figure III: Intra-operative endoscopic picture of the mass

hemangioma (Figure IV). So far, with a follow-up duration of more than 12 months, no signs of recurrence have been observed.

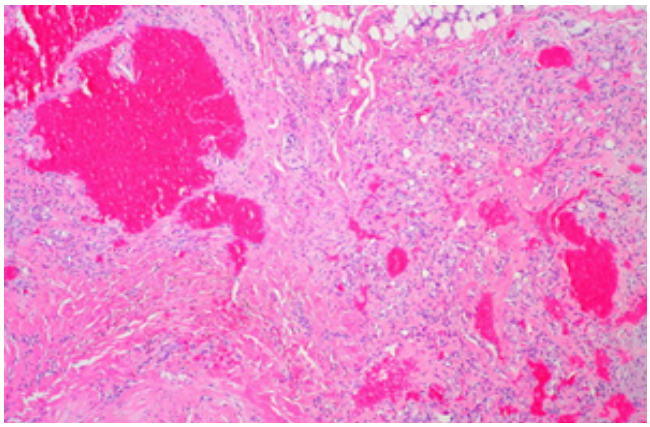

Figure IV: Histopathology showing the lesion composed of thick and thin-walled vascular channels compatible with cavernous hemangioma

DISCUSSION

Haemangiomas are benign vascular tumours composed of newly formed vessels with endothelial lining. Head and neck is a common site for hemangioma. Hemangioma of the nasal cavity and sinuses is extremely rare with only a few isolated case reports describing their relation to the maxillary sinus with a bone eroding property.[4] Depending on the dominant vessel size on microscopy, they are of three types:capillary, cavernous and mixed. Capillary type is the most common type of haemangioma in the nasal cavity, composed of capillary sized vessels, occurring more commonly in the nasal septum or vestibule and being more common in children.Cavernous hemangiomas on the other

hand arisemore commonly from the lateral wall of the nasal cavity, appear at around the fourth decade of life and contain large endothelium-lined vascular spaces.[5]

They are often asymptomatic butcan present with reddish, polypoidalor sessile mass causing a nasal obstruction or recurrent epistaxis, especially in women.Other presenting symptoms could be rhinorrhea, facial swelling and bulging of the eye.[2] In the nasal cavity and paranasal sinuses, they arise from the inferior turbinate, vomer, lamina perpendicularis, osethmoidalis and maxillary sinus.[6] Thrombi within these vascular spaces can occasionally calcify and be identified in CT as phleboliths. There might be an erosion of the medial wall of the maxilla, widening of the osteomeatalcomplex, and involvement of adjacent sinuses like the ethmoids.

Cavernous hemangiomas are quiescent or slow-growing benign lesions and can have a compressive mass effect.[7] For a definitive diagnosis to be made, histological confirmation of the surgical specimen is a must. However, it is not an easy task as severe bleeding may occur. Imaging and routine blood investigations should be performed prior to any attempt of biopsy.

Differential diagnoses of sinonasal cavernous haemangiomas include long-standing sinonasal polyps, mucocele and inverted papillomas, and if there is associated bone destruction it may simulate a malignant tumour.[8,9] Angiography may provide an idea regarding vascularity as it is both diagnostic and therapeutic.

Cavernous hemangiomas may regress spontaneously. In symptomatic patients, treatment is primarily surgical, ranging from complete excision to local resection. The approaches could be open, endoscopic or combined depending on the situation and extent of the lesion.[10] LASER excision and steroid therapy have no role in the treatment of these lesions. In our patient, the minimally invasive transnasal endoscopic technique had proven to be effective. Tumour was completely removed and the patient on herone-year follow-up hasn't had any recurrence.

CONCLUSION

Cavernous haemangiomais an uncommon benign entity of the paranasal sinuses. Owing to the contrast-enhancing and bone eroding property on imaging, it can mislead to malignant pathology. A pre-operative embolizationfollowed by endoscopic excision can confer a complete cure.

REFERENCES

1. Panbude SN, Guha AH, Mahajan A, Sable NP, Arya S. Cavernous hemangioma of the nasal cavity: mimicking as the inverted papilloma. Int J Otorhinolaryngol Head Neck Surg. 2017;3(2):423.
2. Hamdan A latif, 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.
3. Das SK, Saha S, Ghosh LM, Bhowmick A. Haemangioma of maxillary sinus. Indian J Otolaryngol Head Neck Surg. 2001;53(1):65-67.
4. Kim HJ, Jung Hee Kim, Jae Hyoung Kim, Eui Gee Hwang. Bone erosion caused by sinonasal cavernous hemangioma: CT findings in two patients. Am J Neuroradiol. 1995;16(5):1176-1178.
5. Eivazi B, Ardelean M, Bäumler W, et al. Update on hemangiomas and vascular malformations of the head and neck. Eur Arch Oto-Rhino-Laryngology. 2009;266(2):187-197.
6. Jammal H, Barakat F, Hadi U. Maxillary sinus cavernous hemangioma: A rare entity. Acta Otolaryngol. 2004;124(3):331-333.
7. 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.
8. Kramer D, Durham JS, Sheehan F, Thomson T. Sinonasal undifferentiated carcinoma: Case series and systematic review of the literature. J Otolaryngol. 2004;33(1):32-36.
9. Penezić A, Čupić H, Baudoin T. Mixed Type Haemangioma of the Inferior Turbinate: A Rare Cause of Epistaxis. Indian J Otolaryngol Head Neck Surg. 2019;71:1695-1698.
10. Salimath K, Ramakrishnan N, Galagali J. A Rare Presentation of Cavernous Hemangioma of Both Inferior Turbinates. Bengal J Otolaryngol Head Neck Surg. 2017;25(2):111-114.