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Case Report

Cavernous haemangioma of right nasal cavity: a case report

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ABSTRACT

Hemangiomas are common, benign tumors which arise from the proliferation of endothelial cells surrounding blood-filled cavities. Cavernous hemangioma of the nasal cavity is a rare clinical entity but it should be considered in the differential diagnosis of the nasopharyngeal mass and in cases with epistaxis. This lesion is usually found on the lateral wall of nasal cavity and are more common in elderly with no sex preponderance. We present a case of cavernous hemangioma arising from the sphenopalatine area in a 54-year-old female. The tumour was successfully excised by trans-nasal endoscopic approach with pre-operative surgical embolization.

Keywords: Cavernous hemangioma, Epistaxis, Histopathology, Endoscopic excision, Embolization

INTRODUCTION

Hemangiomas are common, benign tumors which arise from proliferation of endothelial cells surrounding blood-filled cavities. Histologically, there are 3 types of hemangiomas including capillary, cavernous, mixed/ cellular. Capillary hemangioma is composed of capillary sized vessels lined with flattened epithelium separated by collagen stroma while cavernous hemangioma composed of large endothelium-lined vascular spaces and mixed type shows proliferation of thin-walled blood vessels of several lined by endothelium.[1] Although nasal hemangiomas are common in head and neck region, it is rarely seen in nasal mucosa.[1] When this tumor arises in nasal cavity, it is predominantly capillary, occur on nasal septum, more commonly in children and affect more female than male. Cavernous hemangiomas on the other hand, are usually found on lateral wall of nasal cavity and are more common in elderly with no sex preponderance.

CASE REPORT

A 54-year-old housekeeper, presented with chief complaint of intermittent bleeding from the right nasal cavity, which was sudden and profuse for the past two months. There were no other associated nasal symptoms such as nasal blockage, anosmia or rhinitis symptoms. Nasal endoscopic examination of the right nasal cavity showed perforated uncinate process with accessory ostium and fleshy mass with prominent vessel seen protruding through posterior fontanelle which bled readily on touch (Figure 1 and 2). The left nasal cavity was normal and both Fossa of Rosenmuller clear and nasopharynx were clear.

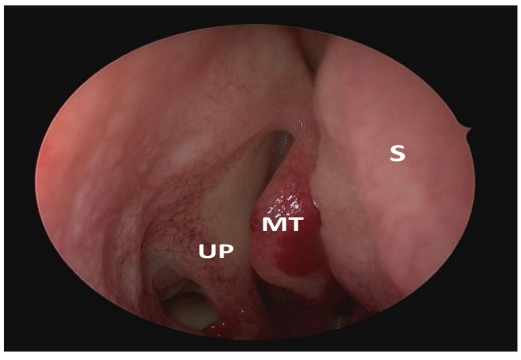

Figure 1: Perforated uncinate process.

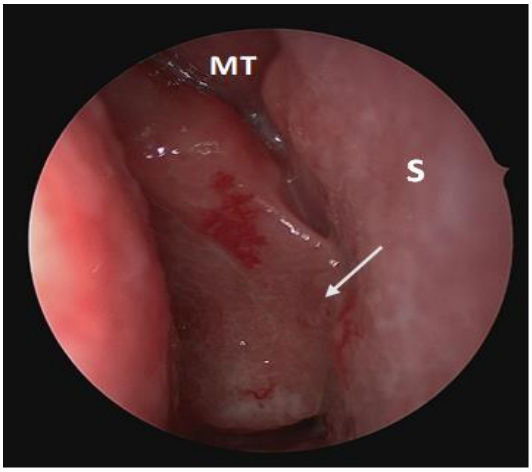

Figure 2: Arrow showed fleshy mass with prominent vessel in the right nasal cavity.
MT- Middle turbinate, UP-Uncinate process, S-Septum.

The patient was suspected to have maxillary sinus squamous cell carcinoma with differential diagnosis of lymphoma or minor salivary gland tumour. Therefore, a punch biopsy was performed in clinic. There was minimal bleeding during this procedure. However, the biopsy was inconclusive. The paranasal sinus computed tomography (CT) scan showed a lobulated soft tissue mass with patchy areas of enhancement in the right maxillary sinus with erosion of medial wall and extends into the nasal cavity. There was also thinning of bony nasal septum. The mass extended into the posterior choanae but no extension into the nasopharynx and does not involve the other paranasal sinuses (Figure 3 A-C). The impression of CT scan findings was right nasal mass with right maxillary extension and bony erosion suggestive of inverted papilloma and differential diagnosis were carcinoma or polyp.

Patient underwent pre-operative polyvinyl alcohol (PVA) embolization of right internal maxillary artery (IMAX) two days prior to surgery as a suspicion of vascular tumour and anticipated bleeding during surgery. Complete en bloc resection of the mass using coblator and bipolar diathermy was achieved by trans nasal endoscopic right medial maxillectomy and the bleeding was controlled with nasal packing which was removed a day later without recurrence of bleeding. The mucosa at attachment site was partly denuded and bone exposed. Intraoperatively, there was a well encapsulated lobulated mass which arise from sphenopalatine area. The mass has multiple broad base attachment and extends medially into nasal cavity (Figure 4 and 5).

Histopathological study of the resected specimen showed dilated blood vessels lined by flattened endothelial cells (Figure 6-8) consistent with a cavernous haemangioma. The patient was followed up regularly and there was no recurrence of the lesion observed at the one year follow up examination.

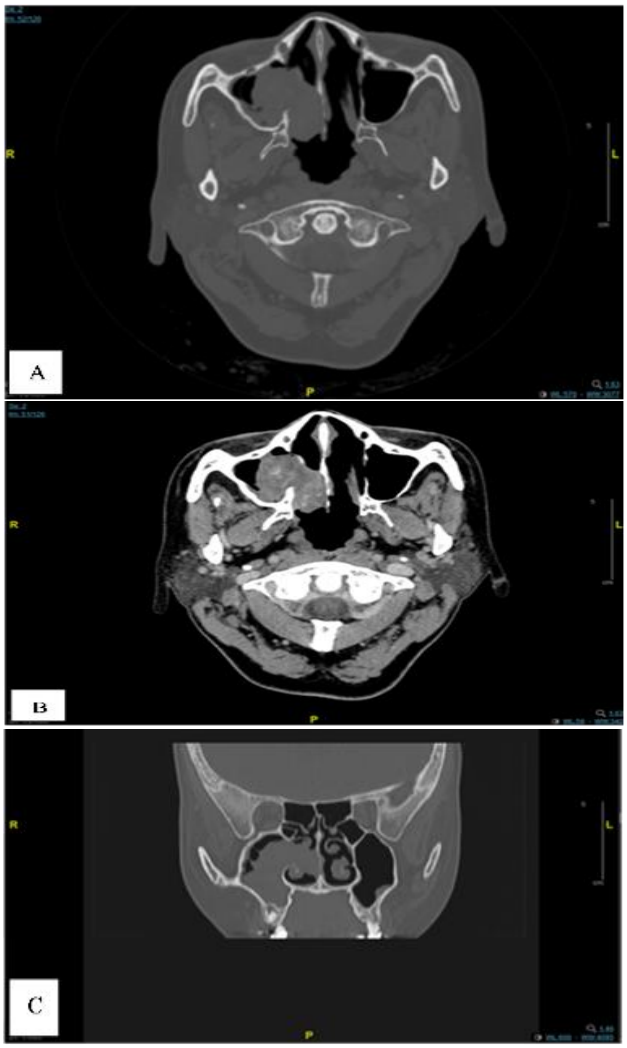

Figure 3 (A-C): Computed tomography scan. Axial cut view non contrasted showing mass in right maxilla extending into the nasal cavity into the choana. The medial wall of maxilla was eroded. This mass showed patchy enhancement with contrast. Coronal cut CT scan shows the mass has widened the maxillary ostium. There was no orbital involvement and the posterior ethmoid sinuses were not opacified. The contralateral sinuses were normal.

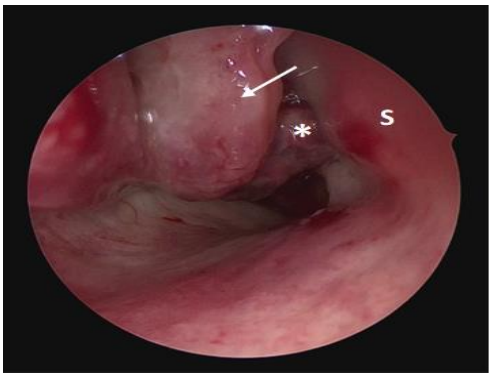

Figure 4: Mass (arrow) extend medially into nasal cavity and also give attachment to nasal septum (*).

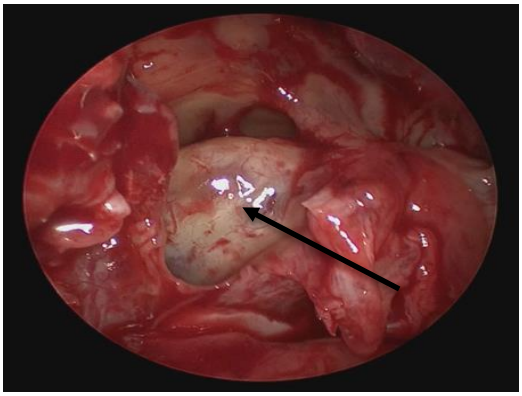

Figure 5: Well encapsulated lobulated mass in the right maxillary sinus (arrow).

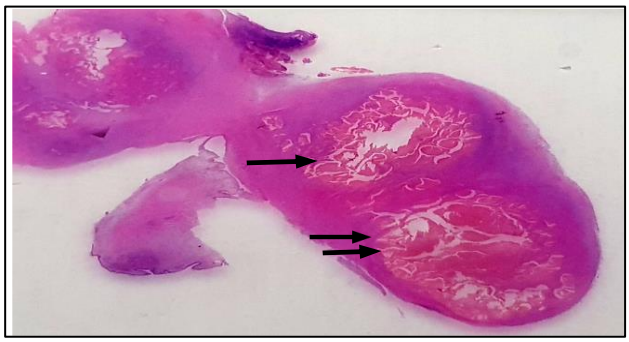

Figure 6: Macroscopic view of slide: Polypoid vascularized lesion, containing multiple cystic spaces filled with blood (arrow).

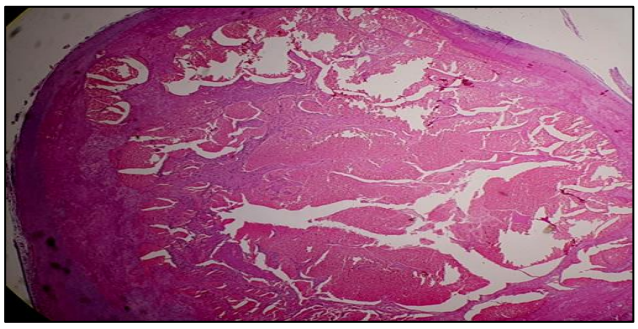

Figure 7: Polypoid lesion, harboring dilated thin-walled blood vessels filled with red blood cells (x20).

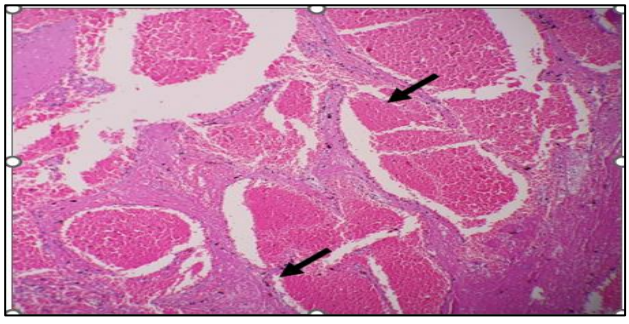

Figure 8: Dilated blood vessels lined by flattened endothelial cells (arrow) (x100).

DISCUSSION

Hemangiomas are common, benign tumors which arise from the proliferation of endothelial cells surrounding blood-filled cavities. Although they are the most common soft tissue tumor in head and neck, they are rare in the nasal cavity and the paranasal sinuses.[2] It can be histologically classified based on the size of the proliferating vascular spaces which are capillary, cavernous, mixed or cellular.[2]

The capillary hemangioma or also known as pyogenic granuloma is more frequently seen where it is usually found on the anterior cartilaginous nasal septum, Little's area or the chondrovomerian junction rather than the cavernous type of hemangioma.[3] The cavernous type of hemangioma is rare and seen in bony septum or lateral nasal wall.[2-4] It can be due to trauma and mostly seen in adulthood over 40 years and without any significant sex difference.[3,4]

The most frequent presenting symptoms for nasal cavernous haemangioma is epistaxis but patient may also present with nasal obstruction, runny nose and epiphora.[5] Less often, patients may also complain of extra nasal symptoms such as facial pain and headache due to sinus obstruction but pain alone is not a characteristic symptom of nasal haemangiomas itself.[5,6]

In the present case, patient presented with unilateral intermittent epistaxis with no previous history of trauma. Macroscopic examination of surgical specimen showed polypoid vascularized lesion, containing multiple cystic spaces filled with blood and dilated blood vessels lined by flattened endothelial cells in microscopic view which is characteristic of cavernous hemangioma.

The differential diagnosis of the nasal haemangiomas includes venous hemangioma, inverted papilloma, lymphoma, and squamous cell carcinoma.[6,7]

The pre- operative biopsy of the tumour for histological confirmation together with computed tomography (CT) or MRI are essential to determine the diagnosis and management planning. Ideally, imaging investigations should be performed prior to any attempt of biopsy. Radiological imaging is also needed to determined anatomical location of the tumour and its extension for treatment planning. If the radiological investigation confirmed a vascular lesion, the next step would be to perform excision under general anesthesia. In this case a punch biopsy was performed in clinic as it did not have the typical vascular mass appearance and lacked large prominent vessels. Fortunately, the mass did not bleed much on punch biopsy. However, it was not conclusive and needed further tissue confirmation. Likewise in our case, Dutta et al reported a case of mucosal cavernous hemangioma of maxillary sinus in which biopsy of the intranasal mass was performed in the office with no significant bleeding during or following the diagnostic

procedure.[8] Kim et al also reported two cases of sinonasal cavernous hemangioma without substantial bleeding post biopsies and surgery.[13] This might be explained by the slow-flow vascular lesion or non- arterial circulation of the cavernous hemangioma.[13] In retrospect, the sudden profuse epistaxis was a major indicator this was a vascular lesion whereas non vascular tumours will present with blood-stained nasal discharge.

The radiographic features of sinonasal cavernous hemangioma have been reported in the CT and MRI literature, but MRI is superior to computed tomography in sinonasal cavernous hemangioma in view of high soft tissue resolution and less artifact.[9] The standard radiological examinations have limited role in determining definitive pre- operative diagnosis.[1] CT scan with contrast feature of cavernous hemangioma is a heterogeneously enhanced soft tissue density mass which is well circumscribed mass. The non-enhancing areas correspond to the areas of necrosis or hemorrhage. The underlying bone is usually normal but may remodeled by adjacent long-standing pressure from the expanding tumour.[10,11] However, there have been reported cases of extensive bone destruction caused by hemangioma of the paranasal sinus. There are two cases of cavernous hemangioma of sphenoidal sinus that caused extensive destruction of the skull base reported by Hayden et al and another two cases of cavernous hemangioma originating in the sinonasal cavity causing adjacent bony erosion were reported by Hyung.[12,13] The nasal turbinates, medial wall of maxillary sinus, maxilloethmoidal plate and wall of orbit were eroded either partially or completely.[13] Likewise, in our case, the tumour led to erosion of the medial wall of right maxillary sinus and minimal erosion of the posterior part of nasal septum. Thus, this nonspecific CT features with associated bone erosion made CT diagnosis quite difficult and hard to differentiate them from the malignant epithelial tumour such as sinonasal squamous cell carcinoma or benign lesion such as inverted papilloma or sinonasal polyp.[13]

According to Itoh et al MR images of cavernous hemangioma show signal intensity of the unclotted blood and they have prolonged T1 and T2 relaxation times, which result in low signal intensity on T1W images and high signal intensity on T2W images.[11] Foci of hypointense signals may represent phleboliths and T1W hyperintensity signal may represent areas of hemorrhage or fat content.

Angiography is warranted if a vascular tumour is suspected. At angiography, foci of pooling of contrast material within the lesion confirm the diagnosis of hemangioma. Other than that, with the aid of trans-arterial embolization using particulate emboli, undue hemorrhage during surgical intervention can be avoided.[12,13] In this current case, the arterial supply of the tumour arose from several branches of the right internal maxillary artery (IMAX). Endovascular embolization using polyvinyl alcohol (PVA) particles was performed 48 hours prior to surgery. According to Duffis et at tumour embolization is best performed 1-7 days prior to surgical resection, which allows for maximal thrombosis of the occluded vessels, optimal tumour necrosis and tumour softening.[14]

Surgical excision of the tumour is the treatment of choice for the treatment of nasal hemangiomas. Various other methods like cryotherapy, corticosteroid treatment, sclerotherapy and laser ablation can be applied. Choice of the surgical approach depends on the nature and exact locations of the tumour, but it should not cause functional and esthetic problems. Many surgical approaches have been suggested including the midfacial degloving, lateral rhinotomy, trans-palatal, trans-antral approach and the Le Fort I osteotomy procedure. The trans-nasal endoscopic approach has been proposed as the technique of choice in cases of intra-nasal hemangioma of the nasal cavity and the paranasal sinuses.[1]

In our case, endoscopic assisted medial maxillectomy was done with complete resection of the tumour associated with minimal intra-operative bleeding. This surgical approach was chosen because it was minimally invasive and proven to be reliable in terms of adequate exposure and visualization of the tumour, complete removal of the tumour and also control of bleeding. Other than that, endoscopic approach has least morbidity, fast recovery time and this tumour is still resectable endoscopically. In contrast to that, this approach has poorer control of bleeding and less visualization. Thus, preoperative optimization of patient's comorbidities, cessation of drugs that may inhibit coagulation and intraoperative controlled of blood pressure, proper patient's positioning (Reverse Trendelenburg), local anesthetic with vasoconstrictor prior to surgery and direct control of feeding vessels using coagulation-suction device plays main roles in control of bleeding during endoscopic skull base surgery.

Nevertheless, there is an alternative for traditional endoscopic approach medial maxillectomy which is intranasal endoscopic prelacrimal recess approach (PLRA) to the maxillary sinus. This surgical approach provides wide access to the walls and recesses of the maxillary sinus especially at the area of prelacrimal recess, and antero-medial-inferior walls of the maxillary sinus[15]. Overall, choosing a reasonable surgical procedure is needed based on the location and extent of the tumour while the nasal function is well protected.

CONCLUSION

In conclusion, pre surgical embolization and trans nasal endoscopic technique was a reliable safe and effective approach for complete hemangioma removal with control of bleeding. The ENT surgeon should be weary of a vascular lesion in patient who presents with recurrent frank epistaxis and cavernous hemangioma should be

considered when the mass appears as a smooth fleshy mass on the lateral wall of an elderly patient.

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