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Ethmoid Sinus Hemangioma - A Case Report and Literature Review

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Abstract

Introduction: Hemangiomas arising in the paranasal sinuses are very rare. In the accessible Pubmed literature 33 cases of sinonasal hemangioma are described out of which only few cases involving ethmoid sinus are described. Even though the lesion is benign in nature, it can be life threatening. Its imaging features are nonspecific, leading to an incorrect preoperative diagnosis in most patients.

Case Report: We present a case of ethmoid sinus cavernous hemangioma in a 66-year-old male. Endoscopic resection of the tumor was performed and the patient is well on follow up. Because of its rarity, literature on paranasal sinus hemangioma is reviewed.

Conclusion: Hemangioma can arise in paranasal sinus and its presentation and imaging findings can be varied. Effective tool for hemostasis is essential to enable a complete excision. Paranasal sinus hemangioma can be managed effectively using endoscope, coblation and microdebrider.

Keywords: Hemangioma, Paranasal Sinus, Ethmoid Sinus, Coblation, Microdebrider.

Introduction

Hemangiomas are benign blood vessel tumours that originate in the vascular tissues of skin, mucosa, bones,

muscles and glands.[1] More than half are found in the head and neck regions. The paranasal sinuses are uncommon sites for haemangiomas[2], especially from the ethmoid sinuses. In the literature many cases of sinonasal hemangioma are described out of which only few cases confined to ethmoid sinus are described. We present a case of ethmoid sinus hemangioma in a 66-year-old male. Because of its rarity, literature on paranasal sinus hemangioma is reviewed.

Case report

A 66-year-old man, presented to us with blood-soaked nasal dressing who had history of profuse nasal bleed from bilateral nasal cavity 5 days ago and was managed in a local hospital with bilateral nasal packing from where he was discharged against medical advice. He also had similar complaints 2 months ago at his home with profuse nasal bleed that stopped spontaneously in around 10 to 15 minutes. He had associated complaints of right sided facial pain and headache for 5 days. He had a history of chronic use of non steroidal anti inflammatory drugs for backache. He had no complaints of nasal obstruction, nasal discharge, allergic symptoms or altered sensation of smell. There was no history of other bleeding tendencies like melena or gum bleed. He had no other comorbidities, and no history of alcohol consumption. Posterior pharyngeal wall was clear and there was no active bleed. Prothrombin time was 16.7 seconds and INR 1.2. Injection Vitamin K 10mg was given intravenously for 3 days twice a day. C-reactive protein (CRP) was 78.3 mg/dL. He was also given intravenous antibiotics.

He was taken to the operation theatre and pack removal was attempted. On partial removal of pack on right side active bleed was noted, immediately he was intubated and packing was done around the endotracheal tube. Left nasal cavity pack was completely removed with no active bleed seen. On endoscopic examination, mucosal change due to long standing nasal pack was noted. Right nasal cavity had active nasal bleed with purulent discharge on removal. On careful examination bleeder was noted adjacent to the middle turbinate. Same was cauterized with suction cautery (Fig. 1a). Anatomy was distorted in the right nasal cavity. Right inferior turbinate was intact with mucosal changes. Right middle turbinate appeared partially resected with few necrotic and bluish black areas within the middle meatus (Fig. 1b). Hemostasis wasachieved and nasal packing done with merocel. Post operative day-2 merocel pack was removed, no bleeding was noted, and follow up endoscopy showed distorted anatomy.

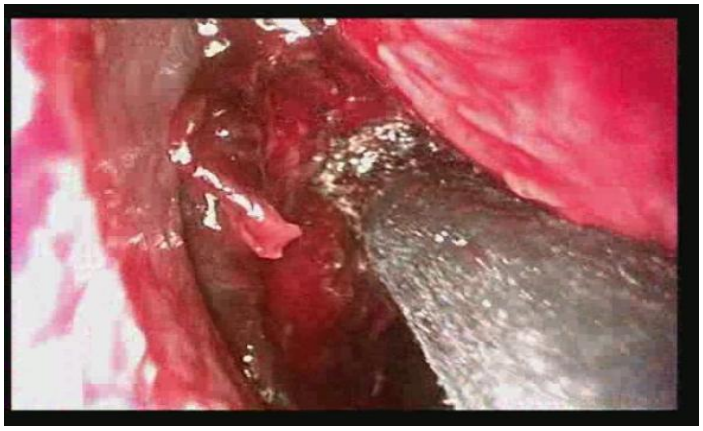

Figure 1a: Bleeding was stopped using suction cautery.

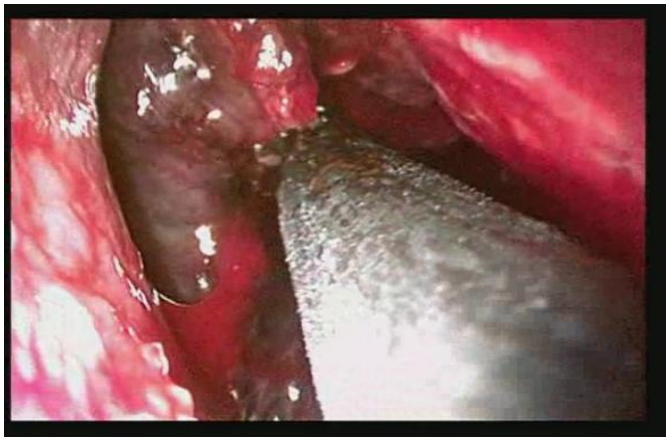

Figure 1b: Dark bluish black lesion seen within right middle meatus.

Later, he underwent contrast enhanced computerized tomography of the paranasal sinus (CT PNS) which showed, soft tissue lesion measuring 32x20x26mm in the ethmoid sinus extending into right frontal sinus and right maxillary sinus with erosion of the right maxillary ostia along with partial destruction of the right middle and superior turbinate (Fig. 2a). Heterogenous contrast enhancement is noted within the lesion with nasal cavity and ethmoid sinus, without calcifications / phlebolith. No evidence of intraorbital extension was noted. Minimal adjacent bony erosion of the right laminar papyracea and adjacent bony nasal septum was noted. The provisional diagnosis of possible neoplastic lesion was considered. Patient underwent MRI scan that showed heterogenous T2 hyper intensity lesion measuring 25x19x23mm in the right nasal cavity partially obliterating the right nasal cavity. The lesion was extending into right frontal recess and was obliterating the maxillary ostia. The lesion was abutting and partially eroding the adjacent lamina papyracea and adjacent bony nasal septum. Mucosal thickening in right sphenoid, maxillary and frontal sinuses were observed, suggestive of sinusitis. The lesion shows iso to hypo intensity in T1 weighted images with homogenous enhancement in post contrast image (Fig 2b). There was no evidence of significant necrosis within the lesion. Left nasal cavity and sinuses were within normal limits.

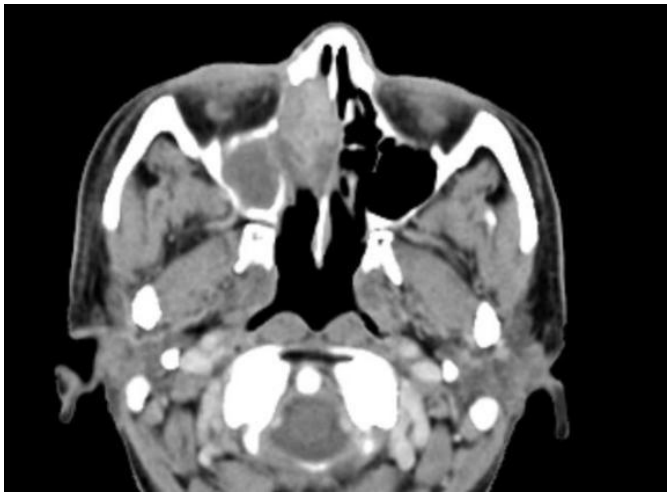

Figure 2a: CT PNS with contrast shows soft tissue lesion in the ethmoid sinus extending into right maxillary sinus with erosion of the right maxillary ostia.

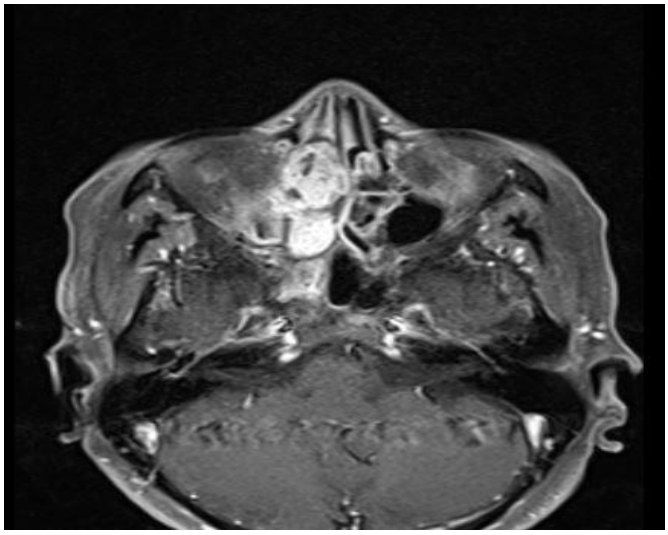

Figure 2b: MRI Contrast T1 weighted image shows homogenously enhancing soft tissue lesion in right nasal cavity and ethmoid sinus.

He then underwent endoscopic microdebrider and coblation assisted complete excision of the mass which was present within the right ethmoid sinus extending up to frontal recess. Intraoperatively it was a very friable mass, torrential bleed was encountered which was controlled with coblation without which it was not possible to remove the mass. Once mass was excised, bleeding came under control. Right side partial middle turbinectomy was also done for the purpose of easier

post operative long-term surveillance. Rest of the sinuses on the right had mucopus within. Fungal ball was found in the right sphenoid sinus which was cleared. 1 Pint of whole blood was transfused intraoperatively. Postoperative period was uneventful. On histopathological examination, dilated and congested thin-walled vessels, with aggregates of endothelial like cells was noted. No nuclear atypia or mitoses was present (Figure 3a). No inflammatory changes were noted. ERG specific for endothelial cells was diffusely expressed in the nuclei of tumor cells (Fig 3b). Histopathology confirmed hemangioma comprising of both capillary and cavernous components.

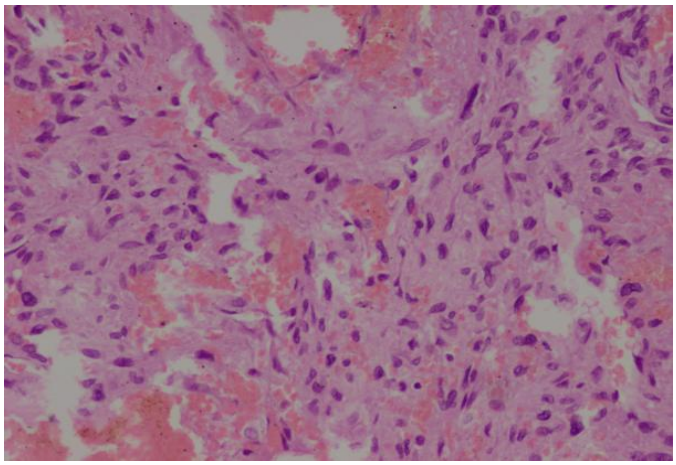

Figure 3a: Histopathology

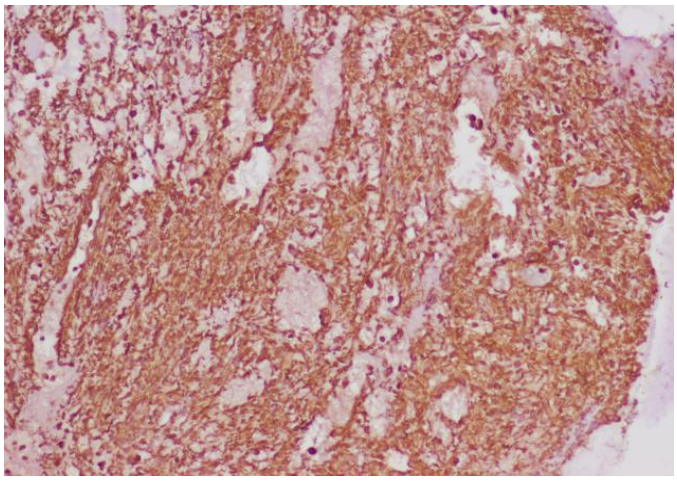

Figure 3b: Histopathology - Immunohistochemistry- ERG positive

The patient was followed-up regularly with endoscopic examination. At the end of 6 weeks, the cavity healed nicely. (Fig 4a, 4b)

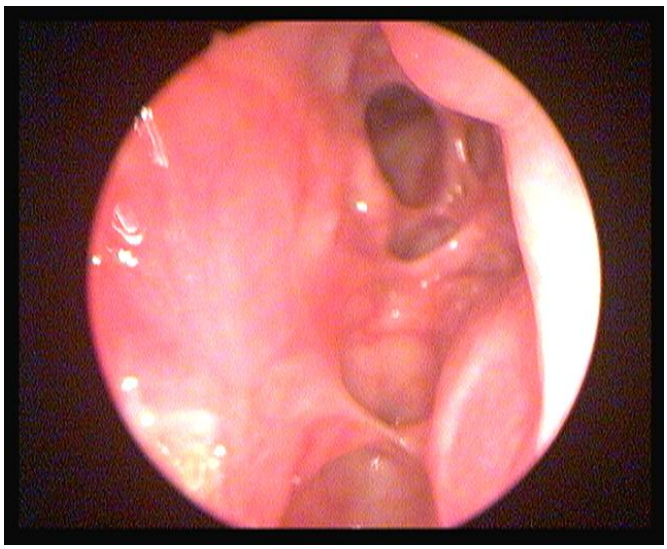

Figure 4a: Post operative endoscopic picture showing frontal sinus and anterior ethmoids

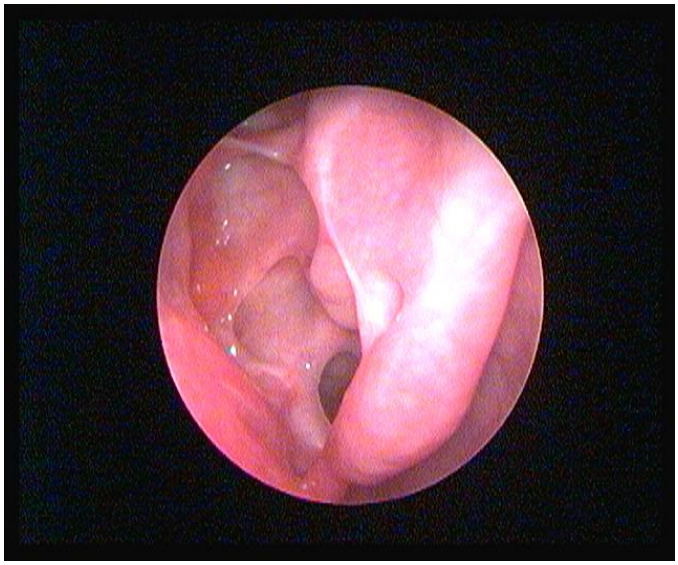

Figure 4b: Post operative endoscopic picture showing ethmoid and sphenoid sinus

Discussion

Hemangiomas are true neoplasm of endothelial cells[3]. However, pathologists now consider these to be hamartomas or hamartomatous malformations[4].

In the literature that was accessible to us, 33[5-29] cases of sinonasal hemangioma are described out of which only

15 cases involving ethmoid sinus are described out of which 8 are confined to ethmoid sinus and nasal cavity like in our case.

Majority of the case reports have presentation of nasal obstruction. In our case, he had presented with severe epistaxis with a history of pack in situ for many days giving a distorted anatomy on endoscopic examination, making the diagnosis difficult. Patient also had a deranged coagulation profile; hence epistaxis was attributed to it. There was no definite mass identified and the discoloration seen within middle meatus was attributed to mucosal changes due to packing which was found to be tumor, that was the cause of bleed. Hence, a case of epistaxis needs further evaluation with imaging once bleeding controlled, especially when endoscopic findings are not clear.

Even imaging findings were misleading. Nasal capillary hemangiomas are homogenous intense enhancing lesions shows iso intensity in T1 weighted images with hypo intensity in T2 weighted image, presence of flow void and intralesional foci of hemorrhage can be seen. However, erosion of the septum and ostia are unusual in hemangioma and common in malignancy. Hence, benign hemangiomas of the sinonasal cavity can cause rather substantial bone erosion, the appearance of which on CT can mislead the radiologist to false diagnosis of a malignant tumor. Various descriptions of hemangioma on imaging have been given .Hemangiomas may be cyst like, or "soap bubble" (sunburst) in appearance, or there may be a radiolucent area with a few scattered striae. On rare occasions they may present as a radiopacity[28]. None of these features is specifically diagnostic on its own, and clinical suspicion should be substantiated by the findings at operation.

Excision of hemangioma can be difficult due to bleeding, in this case the use of coblation was effective in excision of the mass and bleeding was able to be controlled. There are case reports that have reported to use preoperative embolization[10,17] for control of bleeding and have also been effective. The use of coblation during excision of hemangioma of Paranasal sinus has not been described before in literature and is a useful tool in managing such vascular lesions. Coblation has been used in management of oral, oropharyngeal and laryngeal hemangiomas.

Few of the case reports have used external approach[5,7,14,15,23,24] as well as endoscopic for excision and in our case where the tumor was confined to ethmoid sinus and nasal cavity endoscopic approach was adequate.

Histopathologically it was hemangioma comprising of both capillary and cavernous components. The need for histologic diagnosis of cavernous versus capillary hemangioma is not essential because both types are treated by excision[30]. Complete surgical excision should be the goal for hemangiomas arising from the paranasal sinus or nasal cavity. Endoscopic excision with coblation and microdebrider is an effective method in management of ethmoid hemangioma.

Conclusion

Hemangioma can arise in paranasal sinus. Presentation and imaging findings of hemangioma can be varied. Effective tool for hemostasis is essential to enable a complete excision. Paranasal sinus hemangioma can be managed effectively using endoscope, coblation and microdebrider. One should be suspicious of hemangioma in patients who present with severe epistaxis.

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