Case Report

Regurgitated Presentation of Extranasopharyngeal Angiofibroma in Emergency

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ABSTRACT

OBJECTIVE: To add more knowledge about extra-nasopharyngeal angiofibroma presentation.
STUDY DESIGN: Case Report
SETTING: ENT Department of Jaber AL Ahmed Armed Forces Hospital, Kuwait.
PATIENT AND METHODS: An old man of 65 years presented in emergency department with regurgitated angiofibroma of right vallecular region. It was excised from its site of origin with no complications.
RESULTS: Excision was complete and follow up showed no recurrence.
CONCLUSION: Extra-nasopharyngeal angiofibroma is a very rare entity. Complete excision will result in no complication and recurrence.

KEYWORDS: Extra-nasopharyngeal angiofibroma, uncommon disease, emergency presentation.

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INTRODUCTION

The angiofibroma, although a benign tumor but has aggressive nature, most commonly occurs in nasopharynx and due of its higher prevalence in younger age group, it is called juvenile angiofibroma. Rarely it can occur in any age and gender. Its has tendency to spread from its original place of nasopharynx. From nasopharynx it reach to pterygopalatine fossa through sphenopalatine foramen, from pterygopalatine to infratemporal fossa through pterygomaxillary fissure, through inferior orbital fissure in orbit and through superior orbital fissure in Cranium.

There are almost sixty cases recorded in previous literature which have extranasopharyngeal origin. These sites includes inferior turbinate, middle turbinate, nasal septum, maxillary sinus, ethmoidal sinus, infratemporal fossa, cheek and hypopharynx. Histologically irregular vessels are entrapped in fibrous stroma.

Nasopharyngeal angiofibroma cause nasal blockage and episodic epistaxis while other symptoms are related to its extension in neighborhood structures. Facial swelling, proptosis and headache are caused by its extension in pterygopalatine fossa, infratemporal fossa, orbit and cranium. The symptoms of extranasophrayngeal angiofibroma vary according to the site. In the region of interior and middle turbinate its symptom are nasal blockage and hyposmnia. In maxillary sinus as facial swelling, in check as a swelling or mass and in hypopharynx as a foreign body sensation and difficulty in swallowing.

CASE REPORT

In April 2012 a 65 years old patient came in emergency department with history of vomiting and a large mass coming out of his mouth and pedicle of that mass was deep inside the throat. Author was called by his team to manage the case. Patient was conscious and the bilobed mass was tucked to the right angle of mouth (Fig I). He was shifted to ICU and endotracheal tube was passed to keep airway open, avoid choking by falling back of the mass once again inside the throat. After saving the airway attention was given to evaluate the mass. Mass was bilobed, lateral lobe was smaller and pinkish white in color, medical lobe was bigger, firm and reddish pink in color.

Patient was shifted to operation theatre after the routine investigations. The general anesthesia was given (Fig II). Boyles Davis mouth gag passed and pedicle was traced down to right vallecula, Negus artery forcep was applied at the root and excision done. Zero size vicryl knot applied to the root of pedicle. There was a smooth recovery after surgery. The length of the mass was 11.5cm (Fig III), breath of bigger lobe was was 5 cm (Fig IV). After the recovery patient gave the history that he started developing vomiting without reason and this mass popped out of his mouth. He also give history of mild dysphagia on and off. Logically this mass, when in its initial stage was swallowed in the upper part of oesophagus and keep growing there slowly with its pedicle attached in right vallecula. The day he developed vomiting and mass came outside, that obliged him to rush to the hospital. Histopathology report of the mass showed angiofibroma. OPD endoscopic examination after 2 weeks showed complete healing of the surgical wound (Fig V).

FIGURE I: PRESENTATION OF MASS

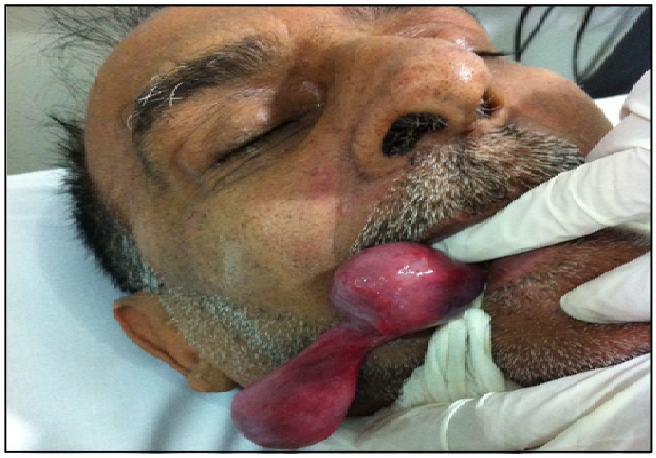


FIGURE II: PATIENT UNDER G/A

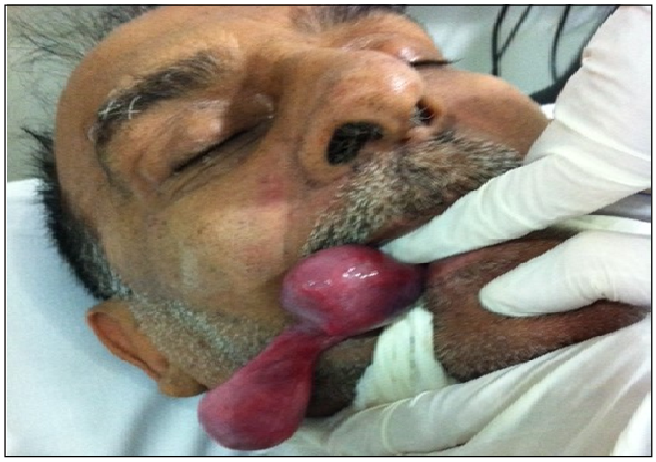
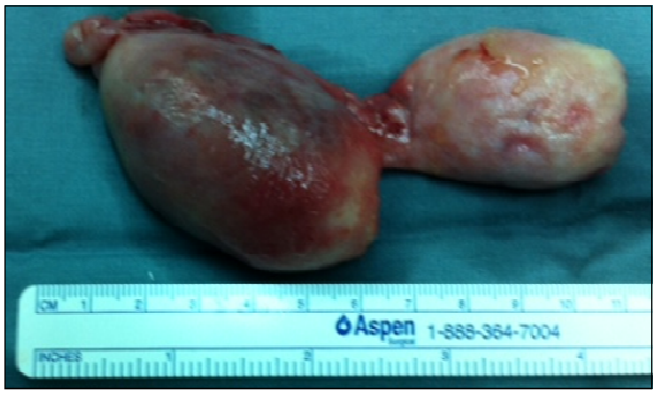


FIGURE III: LENGTH OF THE MASS



RESULTS

After Complete excision from the site of origin, there was no recurrence during the follow up of almost one and half year.

DISCUSSION

The origin of nasopharyngeal angiofibroma is from posterolateral wall of nasopharynx[1] (Trifurcation of root of pterygoid process, horizontal ala of vomer and sphenoid process of palatine bone). It is invasive through foramens and fissures, from nasopharynx it extend into pterygopalatine fossa, to infratemporal fossa, then to orbit and finally to cranium. Angiofibromas are composed of network of vessels and fibrous stroma.[2] Primary extranasopharyngeal angiofibromas are very rare. To date, only sixty cases has been published in the literature, our case number is sixty one. Maxillary sinus is the commonest site of involvement.[3] This is followed by nasal cavity, ethmoid sinus, sphenoid sinus, larynx and pterygomaxillary fissure.[4] One case of posterior wall of hypopharynx has been reported.[5] Angiofibroma has very little or no smooth muscle layer and no internal elastic lamina which may cause severe bleeding when biopsy is taken or have other form of trauma.[6] The diagnosis of classical angiofibroma is clinical and few investigations. Clinically patient is young and complaining of nasal blockage, epistaxis. Endoscopic examination will show the pinkish lobulated mass in nasopharynx. CT scan will confirm the diagnosis for localized or extended angiofibroma. Angiographic examination will further establish the case.[7] Preoperative embolisation is done to reduce the bleeding during surgery.[8] The Major arterial

FIGURE IV: BREADTH OF THE MASS

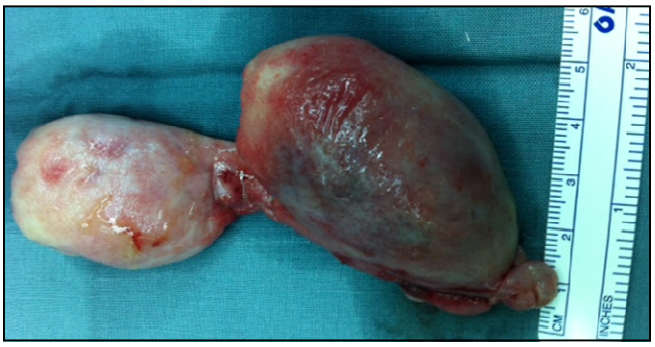
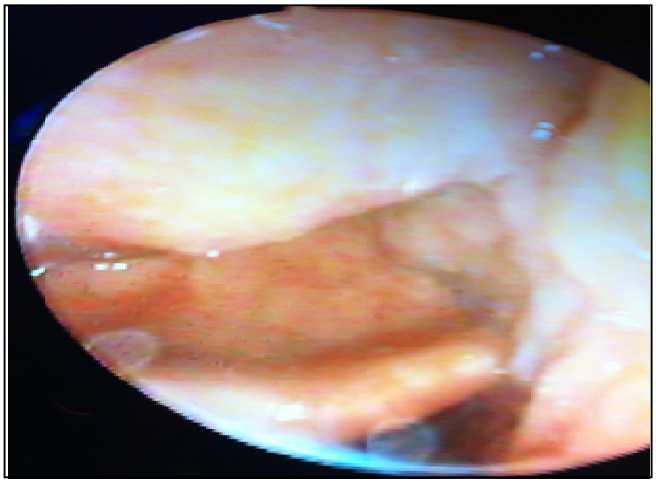


FIGURE V: AFTER 02 WEEKS OF FOLLOW UP (THROUGH ENDOSCOPE)



supply is from the ipsilateral internal maxillary artery but in some cases there is bilateral vascular supply.[9] The fever and facial pain are the two common post embolization sequela of the procedure. Another complication is escape of emboli in intracranial circulation due to reflux or via unrecognized external internal carotid anastomosis.[10] Surgery is the treatment of choice and radiotherapy is reserved for un-resectable & recurrent lesions.[11] Endoscopic removal is possible for small lesions.[12,13,14]

Tumor in the maxillay antrum and anterior part of the nose may be removed through the sublabial approach. Lateral rhinotomy and weberfurguson approaches are limited by its unilaterality and scarring.[15] Mid-facial degloving approach is done for bilaterally extended cases and there is no external scarring.[16] Recurrence can occur both in residual cases & non residual cases.[17] Malignant transformation is also noticed in recurrent cases or in those cases where radiotherapy was used.[18] In our case surgical excision was done because almost all tumor was out and was attached by its pedicle to the site of origin.

CONCLUSION

Extranasopharyngeal angiofibroma are extremely rare and our case is the only case in the literature which was presented in emergency not because of hemorrhage but regurgitated out with vomiting.

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