



CASE REPORT

Extra-nasopharyngeal Angiofibroma: A Rare Case Report

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Abstract

An extra-nasopharyngeal angiofibroma (ENA) is a rare, atypical tumour that arises from sites other than the sphenopalatine foramen. It has a very variable clinical presentation depending on the site of origin, can occur at any age, and does not have a male predominance. This can create difficulties and challenges for doctors tasked with diagnosing the tumour. A rare case of angiofibroma in the left parapharyngeal space is reported as Nasopharyngeal Angiofibroma (NA), which is the most common benign neoplasm of the nasopharynx, but an extremely rare vascular tumor, and represents only 0.05 % of all head and neck tumors. ENAs are even more uncommon. As there are not many cases of this pathology reported in the literature, it becomes extremely difficult to accept this diagnosis except by having a high level of clinical acumen and suspicion.

Keywords: Parapharyngeal Space, Angiofibroma, Extra-nasopharyngeal Angiofibroma, Nasopharyngeal Neoplasms

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Introduction

Nasopharyngeal Angiofibroma (NA) is a rare benign, but locally aggressive vascular tumor that occurs almost exclusively in young teenage boys. However, sometimes angiofibroma can occur in sites other than the sphenopalatine foramen. This is called extra-nasopharyngeal angiofibroma. This is an even rarer tumor with an incidence of 0.05% of all head and neck tumor [1,2], which does not conform to the typical characteristics of torrential epistaxis of NA, and the clinical picture depends on the site of origin. It usually does not recur as its extra-nasopharyngeal origin often facilitates total resection. Histopathology is necessary for accurate diagnosis.

Case Report

A 40-year-old lady presented to the ENT outpatient department of a tertiary care medical college hospital with chief

complaints of difficulty in swallowing and a foreign body sensation in the throat for the past 2 months. Odynophagia was of a gradual onset, with a 2-month duration, and was progressive in nature. It was characterized by difficulty swallowing solids more than liquids, accompanied by a foreign body sensation in the throat. There were complaints of voice change and snoring with no other positive findings.

General examination of the patient did not reveal anything significant.

Local examination revealed a diffuse bulge in the left lateral wall of the oropharynx, which extended to the midline; upper and lower limits could not be visualized (Figure 1). No other abnormal findings. A provisional diagnosis of a left parapharyngeal mass was made with the differential diagnosis of paraganglioma, minor salivary gland tumor, or a nerve sheath tumor.



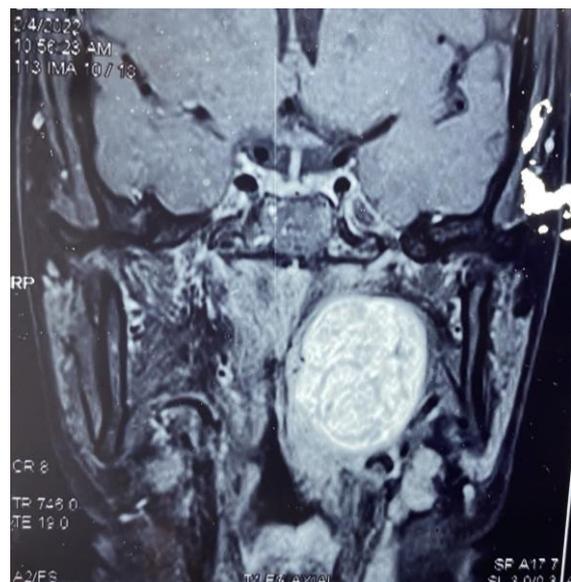
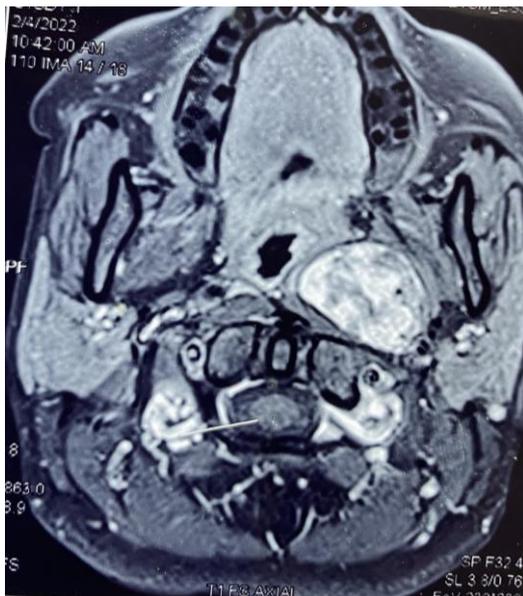
Figure 1. Mass presenting as a bulge in the lateral pharyngeal wall

Diagnostic nasal endoscopy and video laryngeal stroboscopy were both normal. CT angiography revealed a well-defined isodense enhancing lesion in the left parapharyngeal space supplied by the left ascending pharyngeal artery. The left external and internal carotid arteries were pushed laterally along with compression of

the left internal jugular vein. A Carotid Doppler showed a tumor with minimal internal vascularity. MRI and CT scans of the neck revealed an oval-shaped mass measuring 4.5X3.8X2.8 cm in the left side of the neck-parapharyngeal space from the base of the skull to C3 (Figures 2 and 3).



Figure 2. CT showing an isodense parapharyngeal mass



Figures 3a and 3b. MRI picture of the tumour

A surgical decision for excision biopsy under general anaesthesia was made through a transcervical approach. Preoperative embolization was not tried due to the proximity of the tumour to the carotid sheath and due to the small length of the feeding vessel. Under endotracheal intubation, a transcervical submandibular incision was made 2 cm below the mandible on the left side of the neck, extending from the hyoid to the tip of the mastoid. The submandibular gland was gently retracted, and the carotid sheath identified. Branches of Ansa cervicalis and the hypoglossal nerve and the common carotid bifurcation were identified and retracted. On tracing the external carotid artery, a parapharyngeal mass was visualized in the oropharynx with

the help of a tongue depressor (Figure 4). The tumor was found to be ballotable with the capsule just posterior to the carotid. With absolute precaution, the common carotid, internal, and external carotid arteries were retracted and the mass delineated and dissected from surrounding structures and removed in toto with the capsule. The excised tumour specimen was sent to Histopathology and Immunohistochemistry. Histopathology confirmed the diagnosis as Angiofibroma with immunohistochemistry positive for IHC-CD34.

Follow-up was done at 1 year and showed that her symptoms resolved with no recurrence (Figure 5).

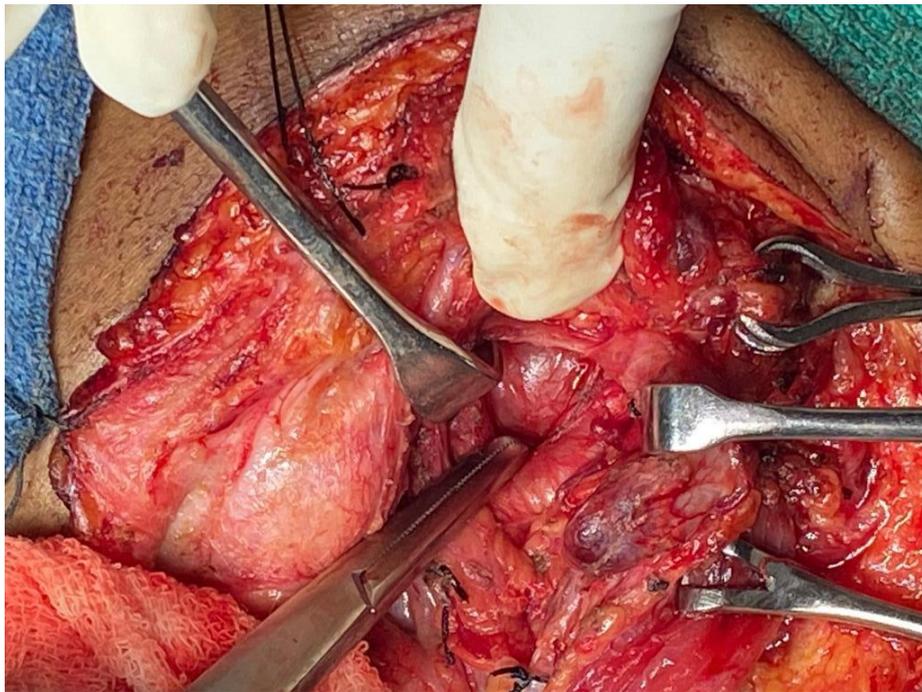


Figure 4. Intra-operative picture of tumour and major vessels



Figure 5. One-year post-op picture showing no recurrence

Discussion

Nasopharyngeal Angiofibroma (NA) is a rare, benign but locally aggressive vascular lesion that occurs almost exclusively in young teenage boys. These lesions typically originate from the sphenopalatine foramen and are present within the nasopharynx. Even though it's benign histologically, it may be locally aggressive with invasion into the bony nasal turbinate, nasal septum, and medial pterygoid lamina. It commonly extends into the nasal cavity, nasopharynx, and pterygopalatine fossa, and can extend into the sphenoid, maxillary, and ethmoid sinuses, and can cause bony destruction. Rarely it spread through the inferior orbital fissure and into the masticator space through the infratemporal fossa. Orbital and intracranial involvement are present in 10 to 37% of cases [3]. The recurrence rate after surgery of nasopharyngeal angiofibroma is around 34% [4].

Extra-Nasopharyngeal

angiofibroma (ENA) is a still rare entity with very few reported cases in the literature [5]. These tumors present with different clinical features, with maximum incidence in females, with a mean age of presentation of 22 years, with a few cases have been reported in children too. The most common site of origin is the maxillary sinus (24.6 %). However, the nasal septum, with invasion of the ethmoid sinus, nasal cavity, larynx, sphenoid sinus, cheek, conjunctiva, oropharynx, retromolar area, middle turbinate, tonsil and inferior turbinate, external nose, hard palate, external ear, lacrimal sac, carotid bifurcation, oesophagus, trachea, facial nerve, middle cranial fossa and infratemporal fossa have been reported in the literature. Devoid of typical clinical and radiological features in all age groups and in females, these lesions arise from various sites, may be less vascularised, and produce

a variety of symptoms depending on the point of origin. Unlike NAs, symptoms of extra-nasopharyngeal angiofibromas are non-specific, like pain, fever, rhinorrhoea, swelling of the cheek, proptosis, headache, progressive nasal obstruction, and occasional epistaxis in a few cases. The main clinical manifestations of ENA were nasal obstruction (80%) and spontaneous rhinorrhagia (60%) [6].

Conclusion

In conclusion, this case gains importance due to the rarity of its presentation and performing the desired surgery in a situation where the plane of dissection involved major anatomical vessels in the neck, it becomes a surgical challenge for every surgeon, as not many are doing such operations on a regular basis to get the desired surgical experience. Though being benign histopathologically, NA and ENA present a challenge to a surgeon due to aggressive growth, intensive intraoperative bleeding, and a high recurrence rate. The clinical manifestations are variable according to their location & harder to diagnose radiologically. HPE is needed for confirmation (acellular connective tissue stroma, matrix of dilated vessels without a muscle layer & lower number of vascular elements). The recurrence rate of ENA is rare because its extra-nasopharyngeal location facilitates total resection. The major hurdles doctors may face are in the accurate diagnosis of this tumour as it is a rare case; each new case may teach us something new about the tumour and its diagnosis and management.

Author's Contribution

Conception and design of the study: KB and NB; Acquisition of data: ASB, NB; Drafting of the article: ASB, NB, KB;

Critical revising: ASB, NB, KB; Final approval: ASB, NB, KB

Conflicts of interest

The authors declare that they do not have conflict of interest.

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Data Availability Statement

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

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