Original Research Paper

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Radio-Diagnosis

VASCULAR CAUSE OF VOICE CHANGE/HOARSENESS OF VOICE - A RARE UNLEASHING CASE REPORT

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ABSTRACT Hemangioma is the most common benign vascular lesion. Laryngeal hemangioma is a rare entity and it is divided into infantile and adult form. Of these two, infantile hemangioma is more common than adult which is most commonly present in the subglottic region which regress spontaneously. Whereas, adult hemangioma requires intervention if large and symptomatic and it is most commonly located in supraglottic and glottic region. This is one such rare presentation in the adult.

KEYWORDS :

INTRODUCTION:

Hemangioma is the most common benign vascular lesion (1), but laryngeal hemangioma is very rare, seen most commonly in children. It is very rare in adults and if present, seen in supraglottic region (2). The incidence of infantile laryngeal hemangioma is 4–5 percent, however, the incidence in adult remains unclear due to the paucity of case reports (3)

Proper clinical history, local examination (including endoscopy) and imaging findings go hand in hand to arrive at proper diagnosis (4).

In this article, we will discuss about the supraglottic hemangioma causing significant air way compromise.

Clinical presentation:

A 45 years old male complaints of voice change x 3 months which is insidious in onset, slowly progressive in nature, hot potato voice with no aggravating/relieving factors with no relieving or aggravating factors. He also had history of difficulty in swallowing for solid foods No h/o difficulty in breathing or noisy breathing or drooling of saliva or odynophagia No h/o fever, cough with expectoration or evening rise of temperature, significant weight loss No h/o nasal discharge, nasal obstruction, post nasal drip No h/o ear pain, ear discharge, aural block, giddiness, facial asymmetry Otherwise, no significant history of prior surgery/ co-morbidities.

Oral cavity and oropharynx examination were normal

Investigations:

On videolaryngoscopy -

- Smooth surfaced, pedunculated swelling arising from left aryepiglottic fold obscuring both the cords and obscuring glottic chink, moves up and down on respiration.
- Left pyriform fossa-not seen
- Right pyriform fossa, arytenoids, epiglottis - normal

Other routine investigations were done and found to be within normal limits.

Imaging findings:

CECT was done for this patient and showed ~ 2.7 x 2.5 x 2.3cm well circumscribed soft tissue density lesion noted arising from the left aryepiglottic fold causing airway narrowing (Figure 1). No evidence of calcification/ invasion into the adjacent structures were noted. On post contrast phase, peripheral nodular enhancement on arterial phase (Figure 2) with progressive enhancement (centripetal filling) on delayed images (Figure 3) were noted. An arterial twig from left superior thyroid artery noted supplying the lesion.

MRI showed T1 hypointense (Figure 4a) and T2/STIR hyperintense (Figure 4b and 5) relatively well-defined circumscribed lesion noted arising from the left aryepiglottic fold causing airway narrowing. No obvious invasion noted.

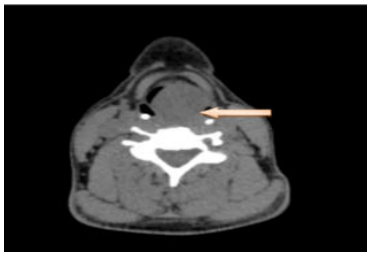

Figure 1: well circumscribed soft tissue density lesion (orange arrow - ⇦) noted arising from the left aryepiglottic fold causing airway narrowing. No evidence of calcification/ invasion into the adjacent structures were noted.

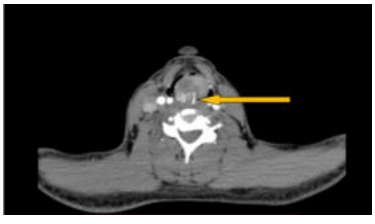

Figure 2: Arterial phase – peripheral nodular enhancement. Arterial twig (Orange arrow- - ⇦) from the left superior thyroid artery noted supplying the lesion

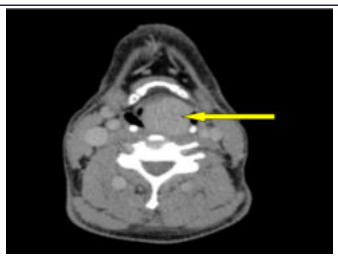

Figure 3: venous phase – progressive enhancement of the lesion (yellow arrow- ⟸) noted

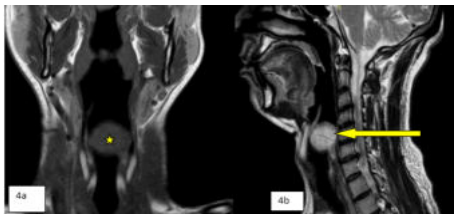

Figure 4a & 4b: 4a – coronal T1W MR image show hypointense well circumscribed lesion (yellow star - ☆); 4b – sagittal T2W MR image shows hyperintense lesion (yellow arrow ⟸) with significant airway narrowing (~80%)

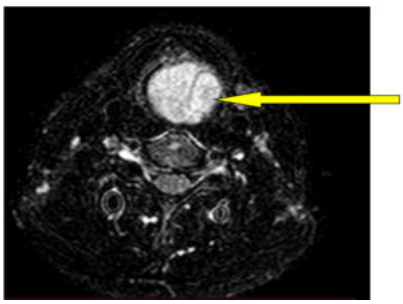

Figure 5: Axial STIR MR images shows hyperintense lesion (yellow arrow- ⟸) in the supraglottic region.

Operative findings:
Patient was taken up for the surgery - Microlaryngeal surgery with supraglottic mass excision and biopsy under local anesthesia on 14/06/2022.

Intraoperative findings:
- Firm globular mass noted arising from the left fold occupying the laryngeal Inlet with broad based pedicle from left aryepiglottic fold and edge of epiglottis. Mucosa of the lesion was pinkish.
- Bilateral false cords, true cords, ventricle were normal
- Subglottis was normal

Histopathological findings:
Nodule show a tissue covered by non-keratinizing stratified squamous epithelium with an attached circumscribed rounded lesion having many lobules of medium sized blood vessels having smooth muscle in the wall admixed with large blood vessels having irregular, dilated lumina and thick muscular wall.

Scanty collagenous stroma is seen.
Perivascular lymphocytic collections are noted.
There is no evidence of atypia / malignancy

Mass from the aryepiglottic fold - Features suggestive of vascular malformation.

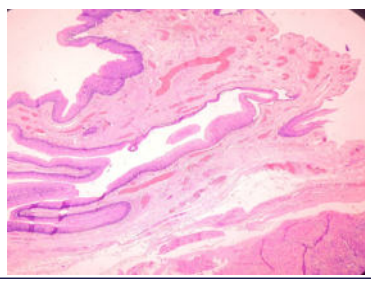

Figure 6a: Scanner view - Nodule show a tissue covered by non-keratinizing stratified squamous epithelium.

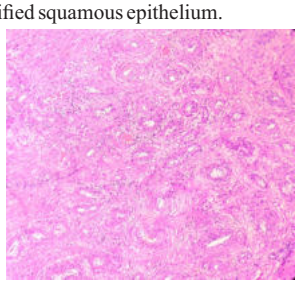

Figure 6b: the nodule has many lobules of medium sized blood vessels having smooth muscle in the wall admixed with large blood vessels having irregular, dilated lumina and thick muscular wall. Scanty collagenous stroma is seen.

Treatment, outcome and follow-up:
Post operatively, patient was started on round the clock steroid according to the and was started on RT Feeds.

On POD-2 ryles tube was removed.
On POD-3 repeat videolaryngoscopy was done and showed edematous left arytenoids with slough, both vocal cords were mobile, bilateral pyriform fossa and epiglottis were normal.

Patient tolerated the oral feeds well. Hence was discharged

Discussion:
Laryngeal hemangioma is a rare entity. It is broadly classified into infantile and adult type. This classification was proposed by Sweetser in 1921 (2). Most common location of infantile type is in subglottic region (5) and adult type is in supraglottic region. Adult type are more often of cavernous form (2).

Cavernous hemangiomas differ from capillary hemangiomas, the latter will have large vascular channels, less well circumscribed and usually deeper in submucosal tissues (6) (7) .

Infantile hemangioma can occur at the age of 2 months upto 30 months(8), usually asymptomatic and regress spontaneously (9). Adult hemangioma can present at any age group and the patient will be symptomatic hence surgical management is often required (8).

The most common reported etiologies are vocal abuse, cigarette smoking, and laryngeal trauma (such as, intubation). The clinical presentation may vary from asymptomatic to voice fatigability, dysphonia, dyspnea (10) and stridor (3) (8). In our case, patient presented with history of dysphonia for the past 3 months.

Clinical history, local examination and radiological imaging can provide us the clue for diagnosis of this lesion.

In our case, on videolaryngoendoscopy, the lesion was pink in color (usually hemangioma appear as a bluish bulge which will clinch the diagnosis), but it was well circumscribed mass arising from the left aryepiglottic fold. Hence possibility of benign etiology was considered clinically.

Contrast enhanced computed tomography (CT) scan and magnetic resonance imaging (MRI) are excellent modalities to evaluate the submucosal spaces, cartilage and extra-laryngeal soft tissues. The staging accuracy of MRI is slightly higher because of more accurate assessment of cartilage involvement and pre-epiglottic, paraglottic extension of tumor. CECT showed well circumscribed soft tissue density lesion noted arising from the left aryepiglottic fold causing airway narrowing with peripheral nodular enhancement on arterial phase with progressive enhancement (centripetal filling) on delayed images were noted.

MRI showed T1 hypointense and T2/STIR hyperintense relatively well-defined circumscribed lesion noted arising from the left aryepiglottic fold causing airway narrowing. No obvious invasion noted.

Hence the possibility of benign etiology – possibly hemangioma was given.

The treatment options of hemangioma include (2):
1. Wait and watch approach if asymptomatic
2. Tumour embolization by introducing artificial embolus in the artery supplying the tumour.
3. Laser micro laryngeal surgery.
4. Co2 laser coblation (6).
5. Sclerosing agent injection.
6. Open surgery.

We proceeded with microlaryngeal surgery for our patient and the histopathological report supported our radiological diagnosis.

Mulliken et Glowacki, based on the histology types, classified the vascular lesions into vascular tumors and vascular malformations (11). Later in 1996, International Society for the Study of Vascular Anomalies (ISSVA) classified the vascular lesions as stratified vascular lesions into vascular malformations and proliferative vascular lesions (tumors) to achieve a uniform classification. In 2014, ISSVA classification was further revised which offers the most widely accepted classification scheme (12,13). Hemangioma comes under vascular tumors.

The practical difficulty during surgery was difficult intubation for this patient.

However, the post operative period was uneventful and he was discharged.

Conclusion:
- Adult hemangioma is rare benign tumor and may present at any age.
- Radiological imaging features helps us to clinch the specific diagnosis.
- Surgical intervention is required if the patient is symptomatic.
- We should always keep in mind of this rare entity.

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