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A CASE OF SUPRAGLOTTIC HEMANGIOMA - A CASE REPORT

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Abstract : A 17year male came with complaints of Cough since three months of age with difficulty in swallowing for the past 3 years and noisy breathing for 10 days. He was treated as bronchial asthma elsewhere. On examination patient was in biphasic stridor. Examination of oropharynx showed pooling of saliva and indirect laryngoscopy was inconclusive. Urgent X ray soft tissue neck lateral view showed edema of supraglottis with prevertebral soft tissue widening. The differential diagnosis according to x ray were acute epiglottitis or retropharyngeal abscess or Vallecular cyst. Emergency tracheostomy was done. CT neck done was inconclusive and suggested biopsy and MRI. Direct laryngoscopy showed soft bluish lobulated mass arising from left arytenoids, left aryepiglottic folds projecting into hypopharynx and oropharynx with normal vocal cords. Biopsy taken was reported as hemangioma. MRI showed vascular mass in left posterolateral hypopharynx supplied by superior thyroid artery. Ligation of superior thyroid artery and Excision of hemangioma was done. The reasons for presenting this case is for the difficulty in initial diagnosis for cause of stridor and rarity of hemangioma larynx presenting in adolescence.

Keyword : Hemangioma, larynx, stridor

INTRODUCTION :

Benign lesions of the larynx are infrequent, about 95% of the benign lesions are papillomas and remaining 5% comprise of oncocytic tumours, pleomorphic adenomas, lymphangiomas, neurofibromas, fibromatosis, paragangliomas, rhabdomyomas and haemangiomas1. Accordingly, haemangioma in the larynx is a very uncommon occurrence, especially in adolescence. Symptoms associated with hemangioma are not diagnostic and mostly are related to the obstruction caused by the lesion. Patients may present with dysphonia, dyspnoea, and dysphagia. Most of the time (in 80% cases) supraglottic region is involved, followed by glottis and subglottis1. A thorough history and complete examination are imperative for diagnosis; however it must be confirmed by radiological imaging and biopsy. Although a biopsy would result in bleeding, differentiating a benign from malignant neoplasm is crucial, as the latter is associated with significant morbidity and mortality. We present a case report of a rare benign supraglottic haemangioma in an adolescent male, who was treated surgically without any complications.

HISTORY:

A 17year old male came with complaints of cough since three months of age associated with difficulty in swallowing for the past 3 years and noisy breathing for 10 days. He had chronic cough since 3 months of age, intermittent and relieved by medication. The Difficulty in swallowing was since three years, initially for solids, now even for his own saliva for the past ten days. He had noisy breathing since 10 days for which he was treated as bronchial asthma elsewhere and got admitted under medical unit and ENT opinion was sought by them. There was no significant past medical history such as previous ENT surgery or intubation and also no history of trauma. On examination patient was in biphasic stridor. He was leaning forward and panting looking emaciated. Examination of oropharynx showed pooling of saliva, occasionally spitting blood stained saliva. Indirect laryngoscopy could not be done. Emergency tracheostomy was done. X ray soft tissue neck lateral view (figure. 1) showed soft tissue opacity in region of epiglottis and prevertebral soft tissue region. X ray offered a differential diagnosis of acute epiglottitis, retropharyngeal abscess or vallecular cyst.

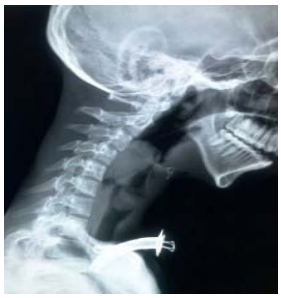

figure. 1

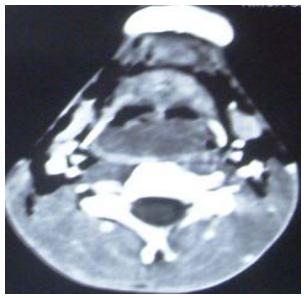

figure. 2

CT neck with contrast (figure. 2) showed evidence of well defined hypodense lesion measuring 43 x 21 mm from C3 to C6 oropharynx involving left aryepiglottic fold & left pyriform fossa with no immediate contrast enhancement suggestive of soft tissue mass in oropharynx and laryngopharynx and advised MRI and biopsy. Direct laryngoscopy under general anesthesia through tracheostomy tube showed (figure. 3) soft bluish lobulated mass arising from left arytenoids, left aryepiglottic folds projecting into hypopharynx and oropharynx. Both vocal cord appeared normal. On probing it seemed to be multilobulated with broad sessile attachment prolapsing into larynx. Guarded biopsy was taken sent for histopathological examination.

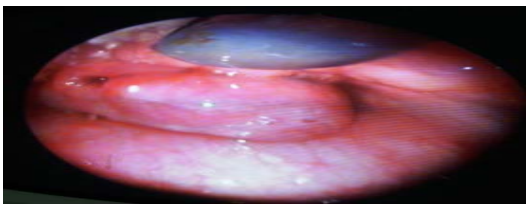

figure. 3

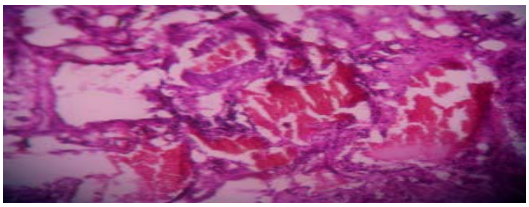

figure. 4

Histopathology (figure. 4) showed stratified squamous epithelium with underlying subepithelium showing numerous dilated blood vessels of varying sizes, some of them containing RBCs separated by thin fibrous stroma suggeative of hemangioma. MRA (figure. 5) showed large vascular mass in left posterolateral wall of hypopharynx extending to left aryepiglottic fold supplied from branches of left superior thyroid artery.

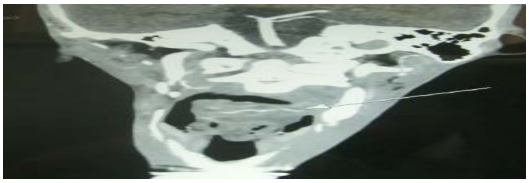

figure. 5

figure. 6

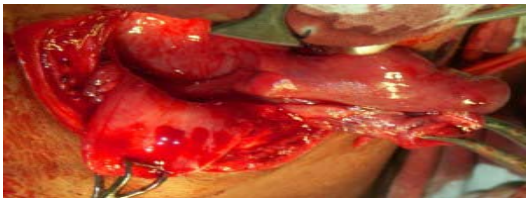

figure. 7

Excision of hemangioma by external approach was done after ligation of superior thyroid artery (figure. 6). Using suprahyoid approach vallecula entered. The mass was attached to vallecular surface of epiglottis on left side, left aryepiglottic fold (figure. 7) with extension into post cricoid region of hypopharynx and was removed in toto (figure. 8 & 9).

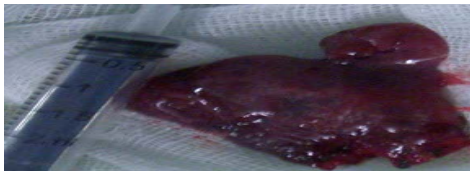

figure. 8

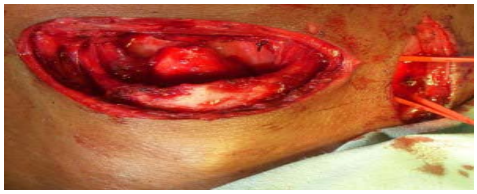

figure. 9

The patient was on tracheostomy and nasogastric tube (figure. 10).

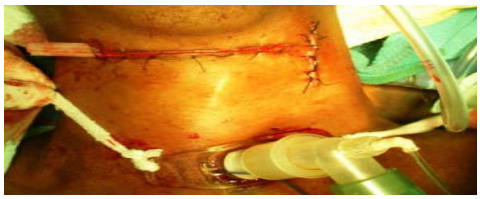

figure. 10

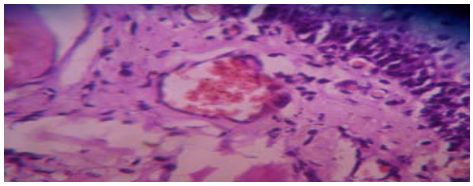

figure. 11

Final biopsy (figure. 11) report showed stratified squamous epithelium overlying fibrocollagenous stroma showing numerous small and dilated spaces lined by flatenned endothelial cells and filled with RBCs. Some of the spaces are filled with eosinophilic proteinaceous fluid and surrounded by focal collection of lymphocytes and suggestive of cavernous hemangioma. On seventh postoperative day nasogastric tube was removed, oral feeding started and spigotting of tracheostomy was done which was subsequently removed and strapping of tracheostome done. Patient was comfortable after that and on followup for past 1 year with no recurrence or symptoms.

DISCUSSION

Incidence of laryngeal haemangioma in adults is unknown due to the scarcity of case reports and case series; in contrast the incidence in infants is 4-5%2. The exact etiology of the origin of hemangiomas is not well understood. Most haemangiomas are congenital and slowly progressive, with 85% of cases noted by 1 year of

age. Pathogenesis is pertinent to the imbalance of positive and negative vasculogenic factors culminating in proliferation of the haemangioma3. Generally, haemangiomas undergo expansion (proliferative phase) in the first 5 months of life, this is followed by regression (involution phase) of the lesion4,5. When the lesion fails to involute, a persistent haemangioma results and forms the platform for uninhibited proliferation. Another theory is that it responds to oestrogen and progesterone hormones, thus increasing in size during pregnancy and decreasing postpartum6. C02 laser excision is the treatment of choice. Hemangiomas of the larynx are generally classified into adult and infantile types. Infantile haemangiomas are usually subglottic and may cause fluctuating respiratory distress and biphasic stridor, especially during periods of venous engorgement. They sometimes accompany cutaneous hemangiomas10. Adult haemangiomas are rare and can be seen at various sites, but are usually glottic or supraglottic. They are more often of cavernous form and cause vague symptoms11,12, such as hoarseness, cough, hemoptysis, dyspnea, and a lump sensation, as in our patient.

Histologically, hemangiomas are composed of large, irregular, blood-filled channels lined with a single layer of endothelial cells between loose fibrous tissue septa of varying thickness. Among hemangiomas, 65% occur in the head and neck region14. Laryngeal hemangiomas are diagnosed primarily by physical examination and history. Doppler ultrasound, computed tomography, technetium imaging, and plain radiographs can play a role in determining the dimensions and extent of hemangiomas15. If the lesion is extensive, angiography and magnetic resonance imaging may be useful in confirming the vascular nature of an adult laryngeal hemangioma as well as in determining its extent. Our patient undergone contrast enhanced CT which did not definitely diagnose the lesion and MRA was conclusive with details of the feeding vessel. There is no well-established treatment protocol for adult laryngeal hemangiomas because only anecdotal case reports or very limited series are available in the medical literature. Injection of corticosteroids or ethanol, cryosurgery, and radiation therapy have been used. For small lesions, excision with microlaryngoscopic techniques can be used. For large lesions, tracheostomy and an open surgical approach may be required.

Laser surgery is thought to be relatively effective and less invasive than surgical removal. CO2 laser excision of the lesion is generally accepted. In limited or pedunculated supraglottic cavernous hemangiomas, CO2 laser vaporization of the lesion with super-pulse modality at 4–8 W is advised12. But CO2 laser cauterization is ineffective in extended lesions and in large vessels with significant bleeding. Extended laryngeal hemangiomas involving the hypopharynx should be approached with staged laser surgical procedures to avoid postoperative laryngeal inflammation and edema12. Previously, nd: YAG laser and cryotherapy were also utilized7,8. Lucioni et al. successfully treated 5 out of 6 patients with supraglottic haemangioma via CO2 laser but had one persistent neoplasm9. Recurrence of haemangiomas signifies an incomplete excision. Laser has favourable outcome with superior conservation and restoration of laryngeal function. However, where laser facilities are lacking then surgery is indicated, with the surgical approach being governed by the location and extent of the growth as in our case.

CONCLUSION

In conclusion supraglottic haemangiomas are rarely reported. There was initial difficulty in diagnosing the cause for stridor in an adolescent male. After diagnosis hemangioma was excised with ligation of feeding vessel by external approach. On followup the patient is completely disease free.

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