University Journal of Surgery and Surgical Specialities

ISSN 2455-2860 2019, Vol. 5(1)

A RARE CASE OF ARTERIO-VENOUS MALFORMATION OF LARYNX.

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Abstract : Arterio-Venous Malformation of the Larynx is a rare vascular lesion, and a very few reports of such lesions are reported in the medical literature.We report a rare case of A-V malformation located over the left pyriform fossa of larynx in a 22year old female ,who presented with a swelling in the left side of the neck . On videolaryngoscopy,smooth bluish swelling was seen in Left pyriform fossa . surgical Excision was performed without complications. The 2-month postoperative follow-up showed good results with no recurrence

Keyword : Arterio-Venous Malformation,Larynx,pyriform fossa,Vascular lesion

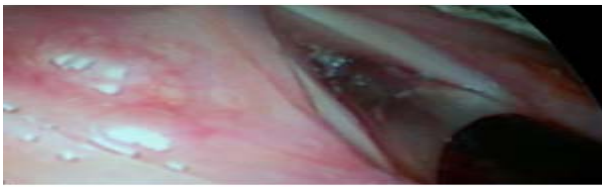
INTRODUCTION

Vascular anomalies are among the most common congenital and neonatal dysmorphogenesis which are separated into hemangiomas and vascular malformations. They can occur in various areas throughout the body with 60% being located in head and neck. True mechanism of the pathogenesis of vascular anomalies is still unclear.

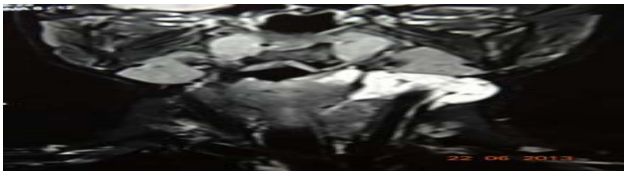
CASE SUMMARY-

22 years old female patient came with complaints of gradually progressive swelling in left side of neck since childhood . This was not associated with any difficulty in swallowing / change in voice/ hematemesis/hemoptysis or respiratory distress.clinically , swelling of about 4x4 cm present at level of thyroid cartilage on the left side extending to anterior border of sternocleidomastoid.It was soft, compressible,non transilluminant ,fluctuant,non tender .

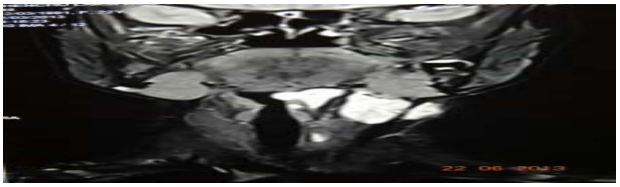
On General Examination
Moderately built and nourished , alert ,conscious and oriented
Not Anemic /Jaundiced
No cyanois /clubbing .
Temp-98F
PR-78/min
BP-110/70mmof Hg
Respiratory Rate-16/min
Cardiovascular system - S1S2 heard, No Murmur
Respiratory System - Normal Vesicular breath sounds ,No Added sounds
Per Abdomen - soft , No Organomegaly
Central Nervous system - Higher Mental Function -Normal ,No FOcal
Neurological deficit
Skin- Normal
Videolaryngoscopy examination smooth swelling present in left Pyriform
fossa which was bluish in colour.

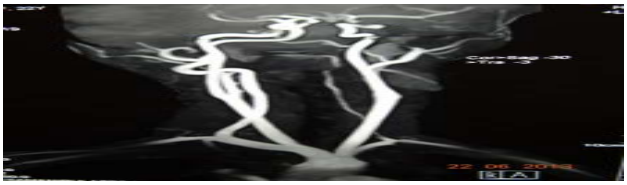


USG finding revealed highly vascular lesion, less likely to be a nerve sheath tumour.
CT neck with contrast shows multi compartmental soft tissue lesion with contrast enhancement in left pyriform fossa,paraglottic space.?Hemangioma ?AV malformation.



MRI neck showed-hyperintense lesion in left pyriform fossa,paraglottic space





Angiography revealed feeding vessels could not be traced.
Hence , Surgery was planned . A horizontal Collar incision at level of mid point of Thyroid cartilage .Skin ,subcutaneous tissue , Deep fascia incised and retracted , bluish mass was seen in the outer surface of thyroid cartilage and extending posteriorly around the posterior border of the thyroid cartilage and occuping the Inner surface of the Thyroid cartilage . The feeding vessels surrounding the vascular lesion was ligated and mass excised in toto.
Post operatively ,specimen sent for HPE which revealed A-V malformation .
4 days after surgery , patient noticed a vascular lesion in ventral surface of tongue on right side. post operative videol aryngoscopy- after one week showed remnant swelling present in the Left pyriform fossa. Hence , CoAblation Assisted Surgical Resection of both the vascular lesions was done .
post operative videolaryngoscopy was normal .
Patient reviewed after 3 months,complete clinical examination and Videolarngoscopy revealed no
evidence of Residual or recurrence of the lesion .
Patient was advised for regular follow up.

Discussion:

Based on endothelial cell characteristics and clinical behaviours of vascular anomalies, Mulliken and Glovacki in 1982 categorized into 2 main categories- Hemangiomas and vascular malformations. Vascular malformations are subdivided according to predominant channel anomaly into either venular, arterial, venous and lymphatic or combinations. Arterial and arterio-venous malformations are high-flow whereas venular, venous and lymphatic malformations are slow-flow. AV malformations of head and neck region include soft tissue and intra-osseous AVM. Soft tissue AVMs used to be called plexiform hemangioma and Arterio-venous fistula whereas intra-osseous AVM used to be called Central hemangioma of the jaw. With profound understanding of the pathogenesis of AVMs, endovascular embolotherapy has become the treatment of choice. Embolization and surgery is often combined for extended cases to improve their facial contour and oral function. Ligation of external carotid artery and/or branches/embolization of feeding arteries with any embolizing agent causes more harm than benefit and should not be used. Once the diagnosis of AVMs is confirmed, an angiography and embolization should be considered the purpose of embolization is to control the growth of AVMs and frequent bleeding the key to embolization is to use sufficient liquid embolizing agents to eradicate the nidus. The currently used liquid agents are ethanol and N-butyl-2-cynoacrylate.Successful embolization is completed when active bleeding has stopped, localized pulsation has disappeared, the lesion becomes lihter in color, the expanded veins in neck return to normal and new bones are formed in cystic zones. For females with AVMs, who are planning to become pregnant, it is best to do embolization before pregnancy, because the hormonal changes during pregnancy may accelerate the progress of AVMs. Ethanol is probably the most effective sclerosing agent that can denature the blood proteins and denude the vascular wall of endothelial cells.however the risks with use of ethanol and the procedure requires skill and experience. ,if the procedure is not adequately performed tissue necrosis and more serious complications such as pulmonary arterial hypertension may occur Soft tissue AVM can be divided into 3 types-infiltrative ,nidus and fistula. For infiltrating AVMs it is suggested to use a mixture of ethanol and contrast at a ratio of 1;1 for embolization where as nidus and AV fistula require absolute alcohol for embolisation.If AVMs of soft tissues affect important anatomic structures with severe disfigurements the most effective treatmentis pre operative embolisation and radical resectionSurgery is indicated when embolization fails or endovascular acesess of the nidus is not possible.surgery is very difficult because of vascularity and lack of distinct margins.surgery should be performed by a experience surgeon with AVMs and ability to reconstruct immediately.It is common for AVMs to recur after surgery,and the surgeon must be ready to reoperate.the goal of surgery must be resect the entire nidus or AVMs will recur.The nidus is very difficult to define because of diffuse feeder vessels and draining vein which don't necessarily have to be resected.In our case preoperative embolisation not done because the feeding vessels could not be identified by CT angiography. LASER are less effective with AVMs and should rarely be used.The only curative treatment of AVMs is the radical resection which is possible only after a pre operative embolisation.

conclusion

Laryngeal A-V malformation is a rare vascular lesion which is usually diagnosed by CT angiography. Preoperative embolisation followed by radical surgical resection is the treatment of choice. In our case , since CT angiography could not trace the feeding vessel of the lesion , only surgical resection was done. Regular follow up is necessary to identify recurrence early.

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An Initiative of The Tamil Nadu Dr. M.G.R. Medical University
University Journal of Surgery and Surgical Specialities