www.jmscr.igmpublication.org
Impact Factor 5.84
Index Copernicus Value: 83.27
ISSN (e)-2347-176x ISSN (p) 2455-0450
crossref DOI: https://dx.doi.org/10.18535/jmscr/v5i4.155

IGM Publication
Journal Of Medical Science And Clinical Research
An Official Publication Of IGM Publication

Arterio Venous Malformation of Tongue – A Case Report

Authors
Dr Avinash Naikwadi[1], Dr Rujuta Rege[2]
[1]Nanavati Hospital, [2]Saveetha Medical College and Hospital

ABSTRACT
AVM of the tongue is a rare cranio-facial vascular abnormality. It occurs because of the failure of complete involution of the fetal capillary bed resulting in development of anomalous connections between arteries and veins. It may be overlooked at birth due to its innocent appearance. Progression of the AVMs is commonly induced by puberty, trauma, and pregnancy. Some forms of treatment, including ligation of arterial feeders, partial excision, incomplete arterial embolization, and laser treatment can trigger progression of quiescent AVM's. Herein, we report a case of 34 – year –old woman who presented with a growing lesion in the floor of the tongue.

INTRODUCTION

Vascular malformations are seen in about 1% of the population. However, many of them do not present for treatment. The first classification was introduced by Glovacki and Mulliken in 1982. This classification was based on the structure and behaviour of these malformations. According to this classification, vascular malformations were divided into arterial, venous, capillary, lymphatic and combined.

Arteriovenous malformations (AVM) is a type of vascular anomaly where there is a shunt between arterial and venous vasculature.

AVM occurs due to the failure of complete involution of fetal capillary bed. This results in the development of anomalous connection between arteries and veins leading to progressive vascular engorgement, venous hypertension, damage of tissue, esthetic problems, and occasionally cardiac decompensation due to high output state.

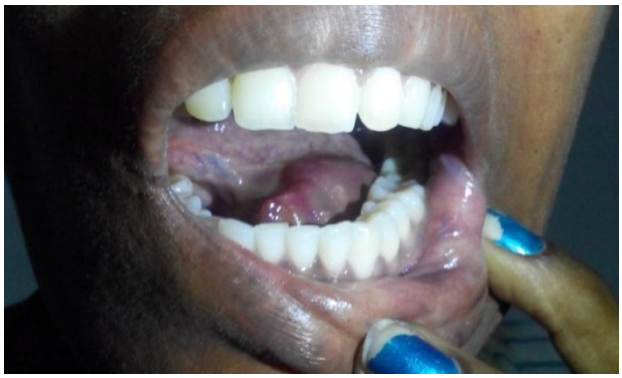
Trauma, puberty and pregnancy can stimulate propagation of AVM. Those induced by trauma usually involve a single vessel. The congenital form of AVM usually involves multiple vessels. Most AVMs are present at birth, but become clinically significant later on in childhood.

AVM can affect any part of the body, but are most commonly seen in the intracranial cavity. Most common extra-cranial location of AVM is the auricle.

CASE REPORT

A 34-year-old was referred to our institution with a swelling present at the floor of the mouth. Her hemoglobin (Hb) at the time of reference was 10.2g%. She gave a history of swelling present underneath the tongue with presence of bleeding. There was no associated pain of the swelling, patient complained of mild difficulty of having solid foods due to the presence of swelling. No other significant history was given by the patient.

Local examination of the floor of the mouth showed presence of bluish red swelling having no tenderness on palpation. (Fig 1 and 2).



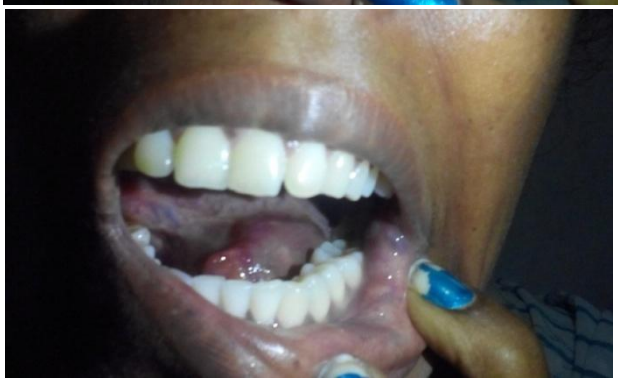
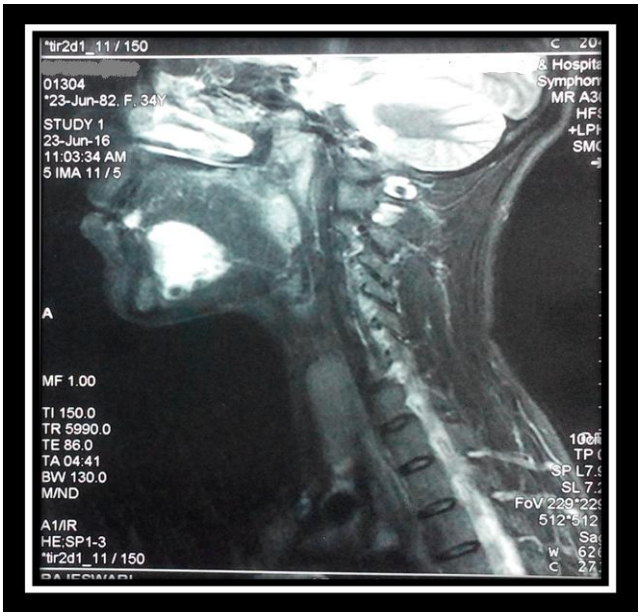


Figure 1 and 2 showing an enlarging bluish red swelling in the floor of the mouth having no tenderness.

MRI done showed Vascular malformation of the floor of the mouth, showing mixed signals and serpiginous nidus of vessels – which could represent mixed vascular malformation of the floor of the mouth. (Fig 3 a , b and Fig 4 a, b).



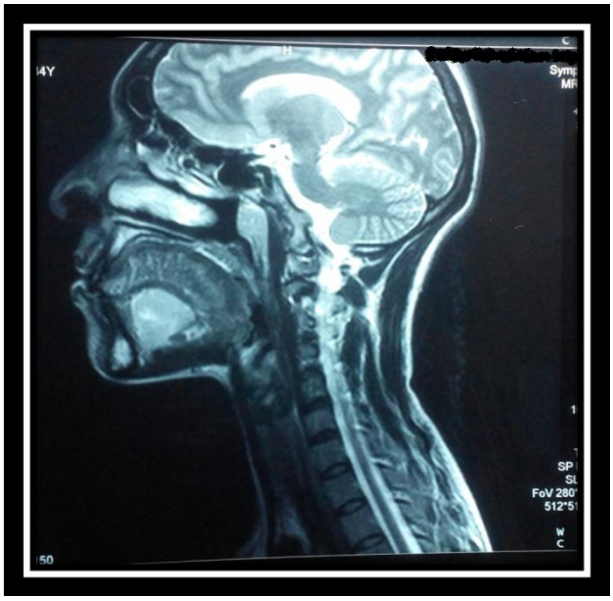


Figure 3 a showing sagittal MR images which show Vascular malformation of the floor of the mouth, showing mixed signals and serpiginous nidus of vessels.

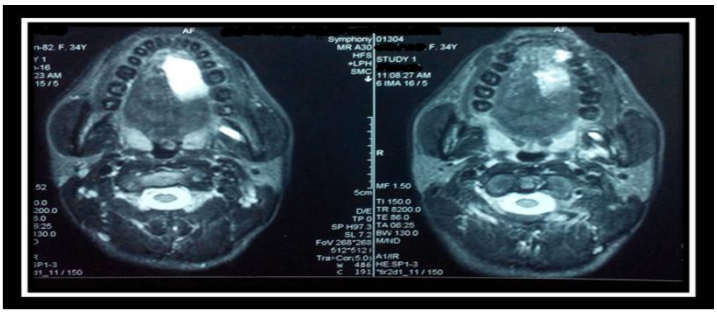
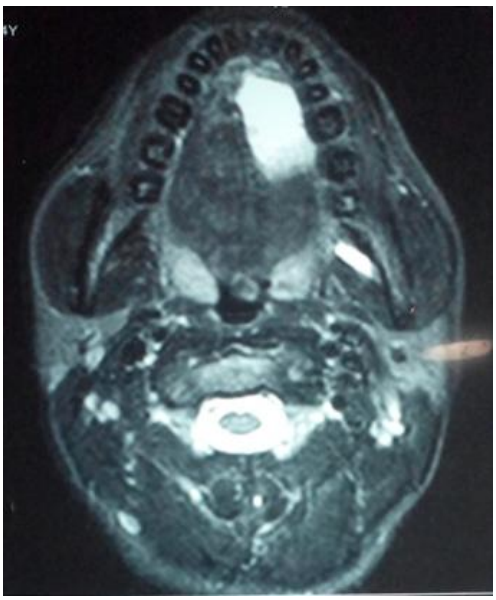


Figure 3 b shows a well defined serpiginous T2 hyperintense lesion is seen in the inferior aspect of tongue predeominantly involving left sided genioglossus.



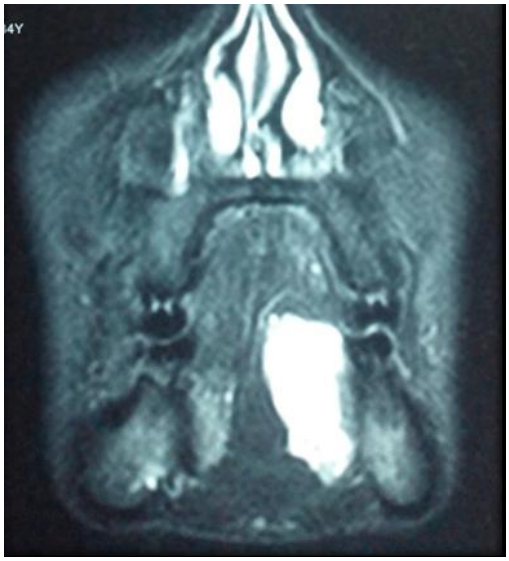


Figure 4 a and b

Patient was taken up for a diagnostic angiogram which showed low flow malformation detected in the floor of the mouth.

Feeder detected was catheterized with with microcatheter and embolized with PVA particles and gel foam.(Fig 5 a, b)

Residual component was accessed with scalp vein set and embolized with 20% 5ml glue, and 90% obliteration of the vascular lesion was obtained. (Fig 6 a)

Post embolization the patient was referred to oral maxilofacial surgery department for removal of the AVM. The department of oral maxillofacial surgery advised re-embolization after 4 weeks, post which surgical excision would be planned.

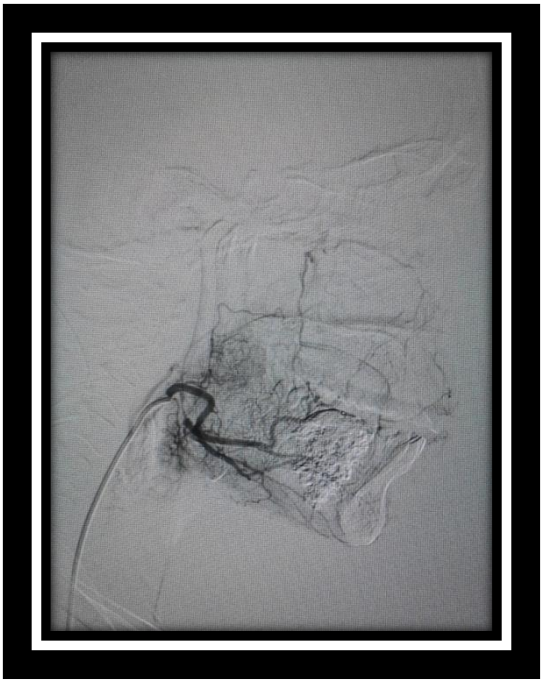


Figure 5 a and b

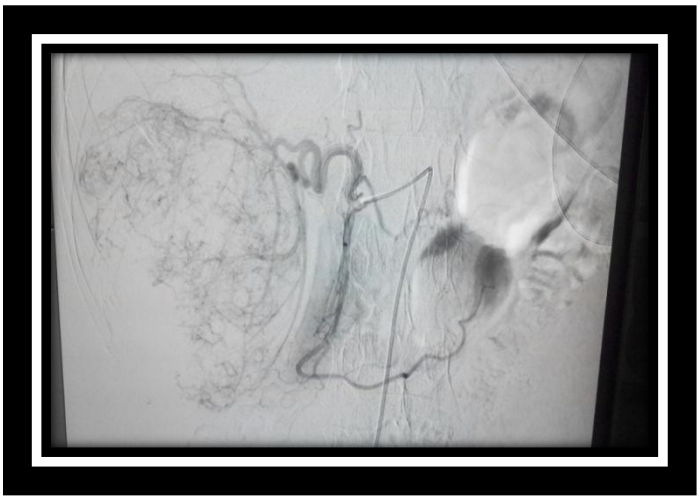
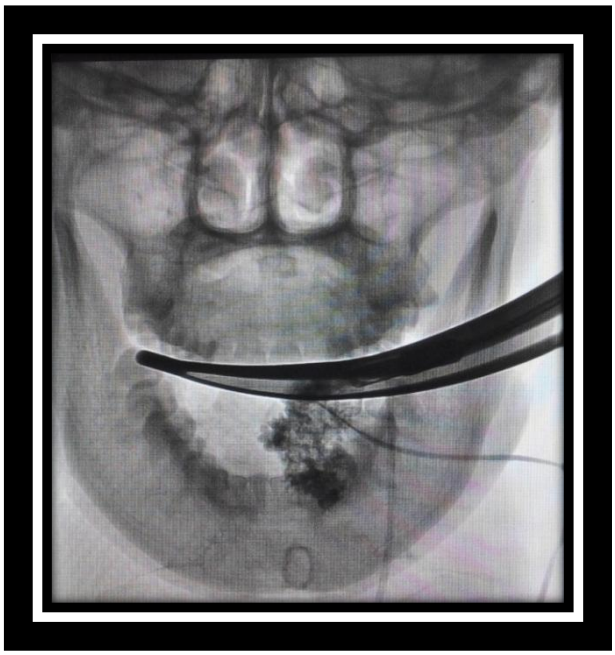
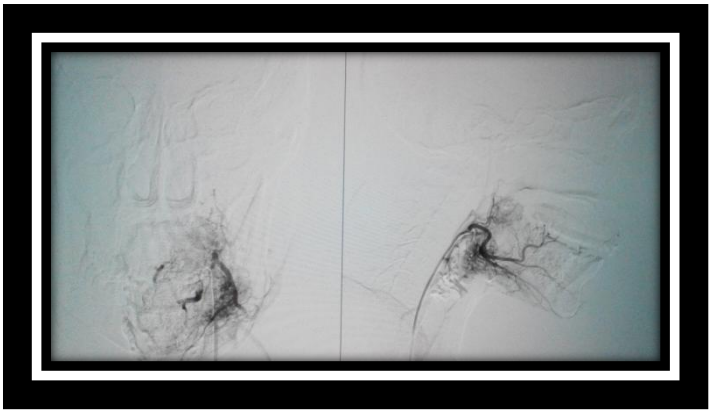
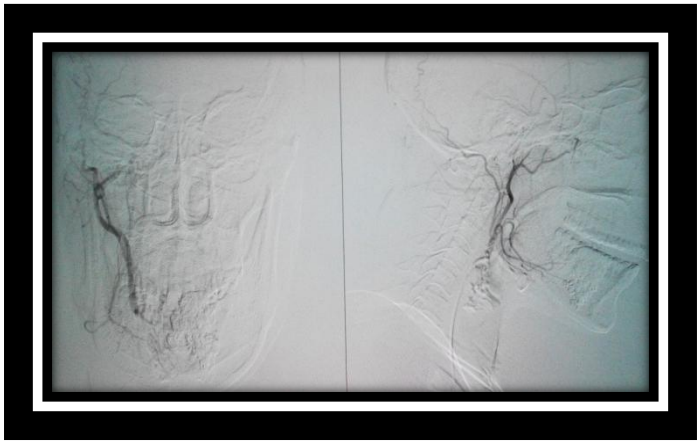


Figure 6 a -AVM of the floor of mouth embolized with PVA particles and Gel foam







Post embolization procedure there is 90 % obliteration of the lesion obtained.

DISCUSSION

AVMs are slow-flow vascular lesions. They comprise of dysmorphic arterial and venous connections without a prevailing capillary bed. They develop, due to the failure of regression of arterio-venous channels in the primitive retiform plexus.

Thus, they are seen in early fetal development itself. The shunting between the high-pressure arterial and low-pressure venous channels account for the clinical picture and progression of the lesion.

Most cases are sporadic, but there are a few inherited syndromes seen along with AVM's.

Mutations in RASA 1 gene , GAP gene show associated congenital malformations along with AVM's.

A defect in ligands or receptors present on endothelial cells can lead to the formation of AVMs.

In hereditary hemorrhagic telangiectasia, AVMs are transmitted in an autosomal dominant fashion.

The incidence of AVMs is equal in males and females. About 40-60% of lesions are visible at birth. 30% of AVMs become clinically apparent during childhood. They may progress in 4 different stages given by The 1990 ISSVA-accepted Schobinger clinical staging used to assess the severity of AVMs.

Stage I lesions –

Asymptomatic having the appearance of a port-wine stain on involuting hemangioma. They are present from birth until adulthood. The presence of a thrill, a bruit or increased warmth may suggest a high flow component in the AVM. Some AVMs may remain in this stage throughout the patient's life.

Stage II –

This phase begins during adulthood. It represents expansion and invasion of deep structures. Progressive dilatation, thinning and fibrosis of arteries and veins is seen histologically. On clinical examination, local temperature is increased and on palpation, a pulse or a thrill can be felt. A murmur may be heard on auscultation.

Stage III –

Grossly it mimics Stage 2. Here involvement of deep destruction occurs with spontaneous necrosis, pain, chronic ulceration and hemorrhage. This stage is seen after years of progressive worsening.

Stage IV –

Characterized by cardiac decompensation and high output failure.

Diagnosis of AVMs is by clinical findings and radiological features. Deferential diagnosis includes other vascular malformations and vascular neoplasms. Plain radiography and CT Scans are not used as a diagnostic tool in these malformations. USG and colour Doppler evaluation helps in the initial assessment however MRI is the investigation of choice. It shows the extent of invasion in these lesions. It provides multiplanar images and differentiates between high and low flow lesions. Existence of flow voids on MRI helps to confirm a fast flow vessel. An arteriogram may be done prior to embolization to know the major feeder vessel. It also gives information regarding the flow characteristics of AVM's and anastomoses. The typical angiographic findings are striking hypertrophy and tortuosity of the feeding vessels. The Nidus of the lesion may differ from large tortuous vessels to multiple small vessels. Collaterals typically have a corkscrew appearance. AVMs are followed up closely at 6 months or yearly interval and treated

only in case of extreme pain, ulceration, bleeding and extensive enlargement.

Treatment of symptomatic AVMs is by palliative embolization whenever combination treatment cannot be performed. For AVMs not amenable to surgery, selective arterial or retrograde venous embolization is used as the first choice of treatment. Different materials can be used for embolization some of which include PVA particles, Gel foam, coils, methyl methacrylate and silicon spheres.

CONCLUSION

AVM's of the tongue are a rare cranio –facial vascular abnormality. Imaging helps in correct diagnosis and immediate management if required in case of profuse bleeding, pain or engorgement in size of the AVM. Supportive measures and palliative care with embolization helps in patient outcome prior to surgical excision.

REFERENCES

1. Watzinger F, Gossweiner S, Wagner A, Richling B, Millesi-Schobel G, Hollmann K. Extensive facial vascular malformations and hemangiomas: a review of the literature and case reports. J Craniomaxillofac Surg 1997;25:335-43.
2. Kaban LB, Mulliken JB. Vascular anomalies of the maxillofacial region. J Oral Maxillofac Surg 1986;44:203-13.
3. Glovacki MJB, Mulliken JB. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. Plast Reconstr Surg 1982;69:412-22.
4. Seccia A, Salgarello M, Farallo E, Falappa PG. Combined radiological and surgical treatment of arteriovenous malformations of the head and neck. Ann Plast Surg 1999;43:359-66.
5. Kohout MP, Hansen M, Pribaz JJ, Mulliken JB. Arteriovenous malformations of the head and neck: natural history and management. Plast Reconstr Surg 1998;102:643-54.
6. Jackson IT, Carreno R, Potparic Z, Hussain K. Hemangiomas, vascular malformations and lymphovenous malformations: classification and methods of treatment. Plast Reconstr Surg 1993;91:1216-30.
7. Malan E, Azzolini A. Congenital arteriovenous malformations of the face and scalp. J Cardiovasc Surg 1968;9:109-40.
8. Chen MT, Horng SY, Yeong EK, Pan QD. Treatment of high-flow vascular malformations in the head and neck with arterial ligation followed by sclerotherapy. Ann Plast Surg 1996;36:147-53.
9. Erdmann MW, Jackson JE, Davies DM, Allison DJ. Multidisciplinary approach to the management of head and neck arteriovenous malformations. Ann R Coll Surg Engl 1995;77:53.