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Research Paper

Arteriovenous Malformation of the Floor of Mouth: A Case Report of Direct Intralesional Sclerotherapy.

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ABSTRACT:
Arteriovenous malformations (AVM) are congenital disorders of vascular structures, causing atypical passage of blood between the arteries and veins that often results in developmental anomalies of the vascular system in prenatal life. Facial arteriovenous malformations (FAVM) are challenging due to its complex vascular supply in the particular region. We present a rare case of a 21-year-old male, who reported to our department with difficulty in speech and restriction in the tongue movements for past 2 years. Clinical examination revealed a palpable diffuse swelling followed by ulceration on the postero-lateral surface of tongue. CT angiography was done, which indicated the hypothesis of a vascular lesion. The histopathologic diagnosis was arterio-venous malformation of the floor of mouth (capillary hemangioma). We treated the lesion using intralesional sclerosing agent and closely followed the patient. The patient had been in a follow up period of 3months and there were no signs of relapse.
KEYWORDS: *Arteriovenous malformation, sclerosing agent, sclerotherapy, ischemic ulcer.*

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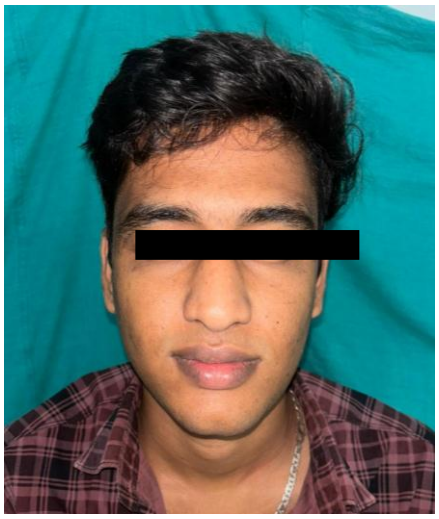
I. INTRODUCTION:

Arteriovenous malformation (AVM) is a condition where there is an abnormal connection between arteries and veins, bypassing the capillary system and these are congenital vascular lesions caused by disturbances in angiogenesis during embryogenesis [1]. The prognosis for individuals with an AVM varies. Untreated AVMs, especially those in the brain, have a risk of bleeding, which can be life-threatening [2-4]. Facial AVMs are dynamic lesions that continuously evolve in their size and complexity in response to previous embolic treatment and with age [5]. The management of AVMs is very difficult, it must be multidisciplinary [6, 7]. Our work is a single case report, on a rare case of localized AVM of floor of mouth extending to ventral surface of tongue.

II. CASE REPORT

A 21 year old male, reported to Department of Oral and Maxillofacial Surgery at SRM Kattankulathur Dental College and Hospital, presenting a lesion on left floor of mouth with restriction in the movement of tongue and speech difficulty for past 2 years. In a local hospital 5 years back, the patient reported a history of swelling accompanying an ulcer in the left postero-lateral aspect of the tongue and floor of mouth not abutting the midline The patient had been suspected for a malignancy and underwent surgical and non-surgical investigations such as incisional biopsy and a computed tomography and assessed to explain the condition as nonmalignant lesion. He neglected any treatment modalities until he presented with pain and progression of the lesion with spontaneous

bleed over the ulcerated growth. No relevant personal or family history could be retrieved during patient interrogation.

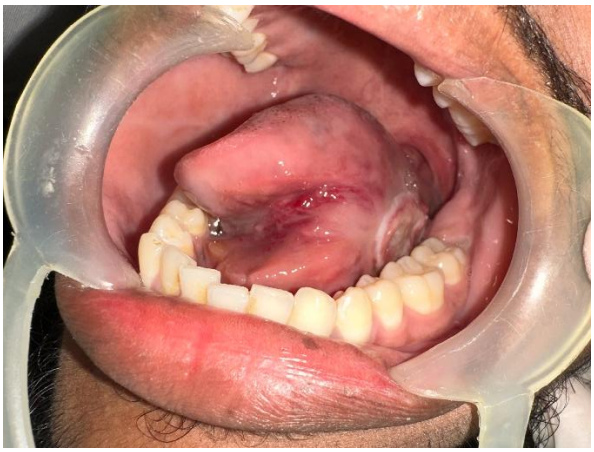


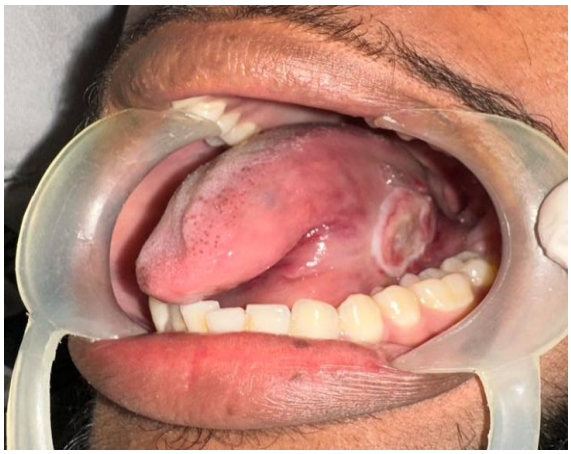
(**fig. 1 :** photo showing mild facial asymmetry on the left side of the face)

Clinical examination revealed minimal facial asymmetry, with an Extraoral swelling in relation to the left side of the face extending to the submandibular region, clear and fluctuant, pulsatile with no prominent vasculature and neck rigidity (fig 1).

Intraorally, there was no restriction in mouth opening, the buccal mucosa and other oral subunits appeared to be normal except the floor of mouth on the left side. On inspection a diffuse swelling along the left side involving the floor of mouth extending into the ventral aspect of tongue, measuring 4x2 cm, with anterior extent 1.5cm behind the tip of tongue and posteriorly upto lingual aspect of 37 region. It appears to be erythematous with no evidence of any discharge over it (fig.2a). An ulcer of 1.5x 2 cm, oval shaped with sloping edges and erythematous base was evident overlying the posterolateral aspect of the swelling (fig. 2b).

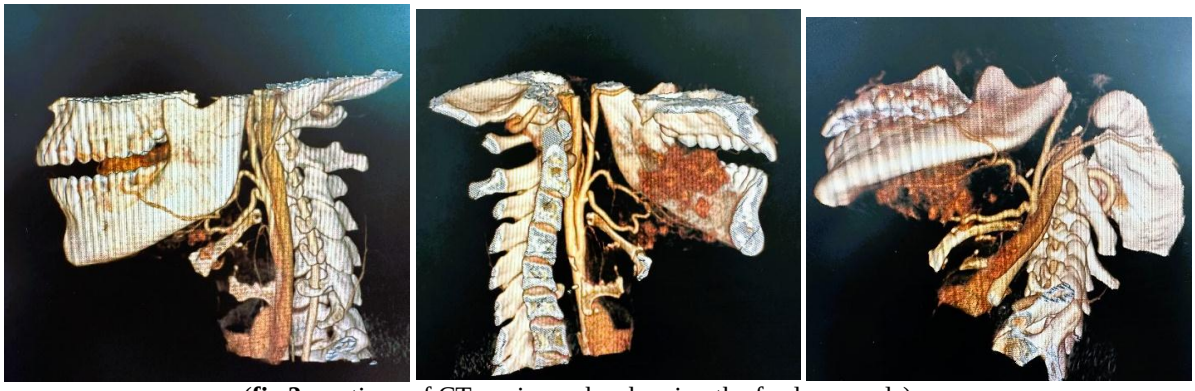
On palpation, all the inspectory findings were confirmed, tenderness present on palpation, with thickened white fibrosed margins, and no indurated base with no evidence of fixity to underlying structures, no active bleed on touch. There was difficulty in speech and restriction the anterior and lateral movements of the tongue with normal deglutition. He also presented with paresthesia of left side tongue.



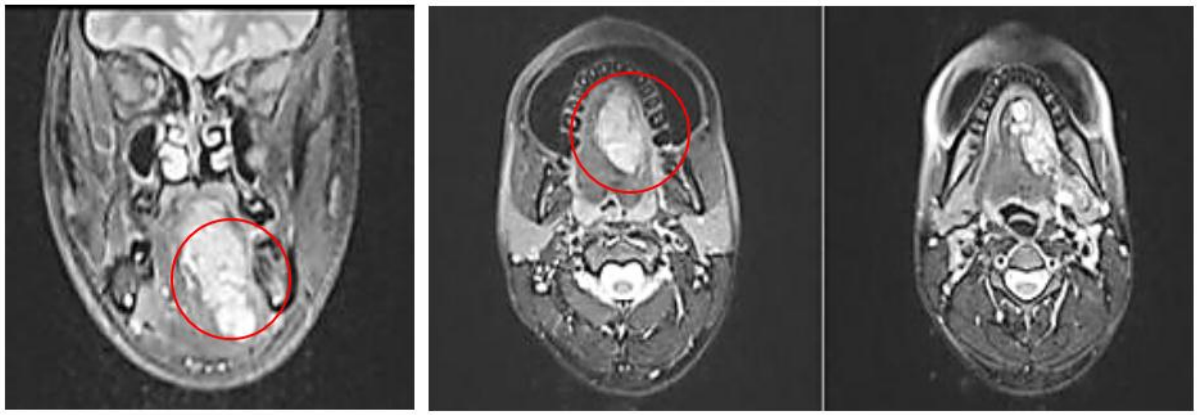


(**fig. 2a and b:** showing the swelling and the ulcerative lesion in the left floor of mouth)

Preoperative laboratory tests done and computed tomographic angiography depicted an ill-defined heterogeneously enhancing lesion in the floor of mouth measuring 4.9x 4.7x 2.7 cm pushing the tongue superiorly and extending the left submandibular and sublingual spaces likely to be a hemangioma (fig.3). Notable features in the region of interest were relative enlargement of the left side compared with the contralateral side. MRI suggested as A well-defined lobulated lesion (T2/STIR heterogeneous hyperintense, T1 iso to hypo intense) measuring 6.1 x 4.8 x 2.7 cm (CCxAPxML) seen at lateral aspect of tongue (fig.4).



(**fig.3:** sections of CT angiography showing the feeder vessels)

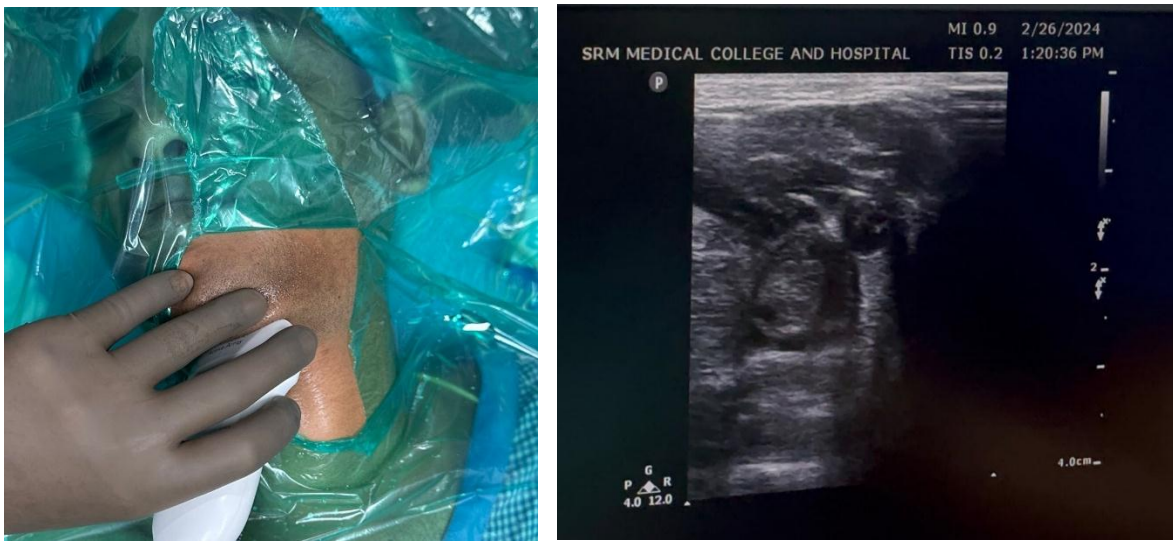


(**fig.4:** sections of MRI showing the location of lesion)

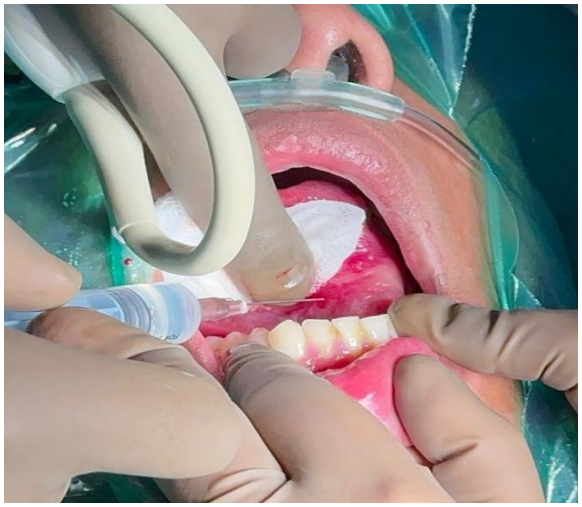
The patient was typed, screened and informed consent was obtained for intralesional alcohol sclerotherapy under deep sedation after identifying the obvious feeder vessels to the region containing the aforementioned region using UGS.

SURGICAL PROCEDURE:

Under deep sedation, standard patient preparation and draping done. Multidisciplinary approach with a team of Oral and Maxillofacial Surgeon, Endovascular surgeon and radiologist were involved for the procedure. USG guided imaging (Extraorally) done to evaluate the site of the sclerosing agent deposition (fig.5). Tortuous course of the feeder vessel visualized and about 4ml of absolute alcohol administered with the guidance of USG.

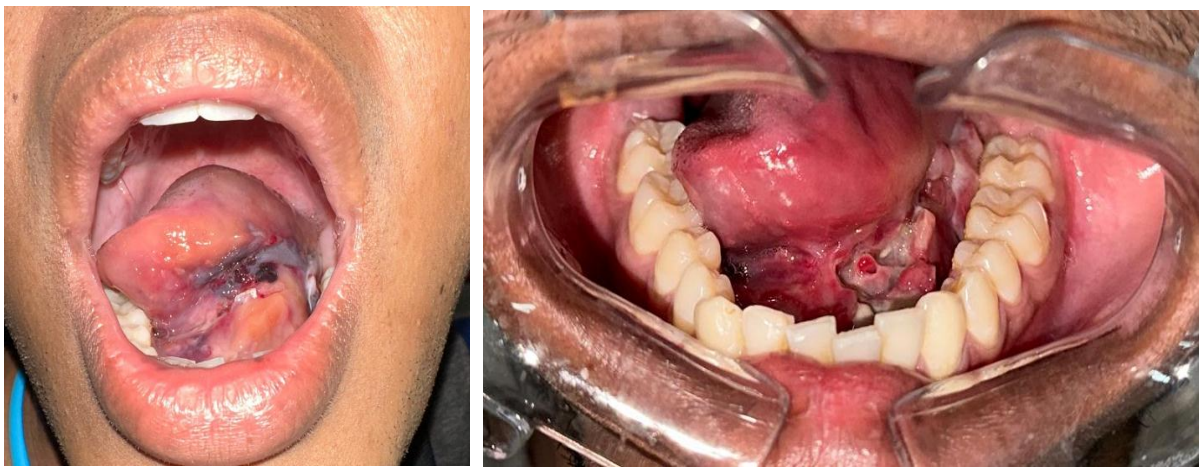


(**fig.5:** loacting feeder vessels with USG guidance)

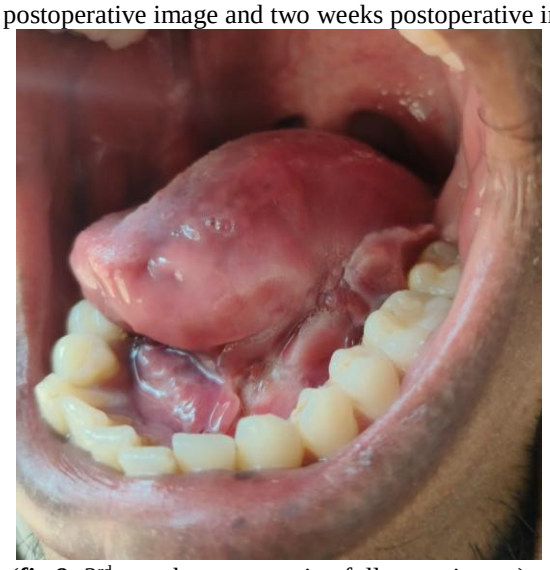


(**fig.6:** intraoral administration of absolute alcohol into the lesion)

Intraorally, tongue reflected 2% lignocaine with adrenaline administered at the site of the lesion and another 4 ml of absolute alcohol administered (fig.6). Using no. 15 blade an incisional biopsy done from the ulcerative region and sent for histopathological evaluation. Localized bleeding spots arrested using electrocautery and hemostasis achieved. Intravenous DEXAMETHASONE mg given as a stat dose.
Post operatively ischemic ulceration was noted on the site of sclerosing agent administration which regressed after a week (fig.7). Patient was followed up for next 3 months (fig.8).



(**fig.7:** immediate postoperative image and two weeks postoperative image)



(**fig.8:** 3rd month postoperative follow up image)

III. DISCUSSION:

Arteriovenous malformation are abnormal connection of vascular structure, between arteries and veins without capillaries in-between [8, 9]. AVMs in the floor of the mouth are rare and present unique challenges due to the rich vascular supply and functional significance of the oral cavity.

Arteriovenous malformations are more commonly presents as congenital abnormalities, others may have later onset in adulthood and lesions of the head and neck are not often encountered in general and vascular surgical practices. The treatment of these lesions has proven difficult due to its high recurrence rate [10, 11].

The etiology of vascular lesions are unknown. The etiopathogenesis of AVMs is still unclear, endothelial dysfunction theory has raised recently due to evidence of endothelial-derived somatic mutations of mitogen activated protein kinase 1 in AVM [12].

Lingual artery, second branch of the external carotid artery, is the main blood supply of the tongue. Venous drainage into the internal jugular vein though the dorsal lingual veins and deep lingual veins. Abnormal communications between the arterial and venous systems shunts a network of surrounding collateral vessels [13]. Here the left facial artery, sublingual and inferior alveolar artery are seen supplying the lesion and they appear prominent.

Our patient did not present with a congenital AVM, but presented with the condition in early phases of adulthood as a localized swelling on the left floor of the mouth which progressed to the current size with no aggravating and relieving factors.

CT angiography (CTA) looks for bone involvement and the vessel and magnetic resonance angiography (MRA) look more commonly on soft tissue involvement. Nevertheless, CT angiography remains gold standard in locating the central feeder arteries, thus providing information for correct diagnosis and treatment planning for embolization or sclerotherapy [14].

A number of adjuvant treatment are used nowadays, such as laser therapy for superficial vessels, and interalesional therapy by injecting a sclerosing agent into the vessels to reduce tissue mass in superficial disease [15], but for a definitive management surgical excision preceded by embolization is the only option.

Since the lesion was localized and visible we advocated sclerosing agent directly into the lesion with USG guidance. After multiple administration of sclerotherapy the size of the lesion regressed with an ischemic ulcer over the lesion which resolved in the due course of time.

IV. CONCLUSION:

Arteriovenous malformations (AVMs) in the floor of the mouth are exceptionally rare and can pose significant diagnostic and therapeutic challenges. This case report illustrates the critical importance of a thorough diagnostic evaluation and a multidisciplinary treatment approach to manage such unusual presentations effectively. Absolute alcohol (ethanol 100%) used to shrink an AV malformation of the floor of mouth was successful. We conclude that AVM in complex locations can still be effectively managed conservatively with interalesional steroids.

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