Case Report

Excision of arteriovenous malformation in an emergency situation - A case report

ABSTRACT

An arteriovenous malformation (AVM) is an infrequent congenital vascular anomaly that can affect the vasculature and involve the endothelium and neighboring cells of any anatomical structure. AVMs are characterized histologically by abnormal AV shunts with atypical interconnecting capillary beds. AVM can cause functional and esthetic issues like face asymmetry, pain, osteolytic changes, and unanticipated hemorrhage or squeeze and tear of the surrounding tissue without causing any symptoms. The literature search yielded limited case reports on AVMs in the facial region. Insufficient diagnosis, limited knowledge, and a lack of literature can lead to severe bleeding and potentially fatal hemorrhagic incidents following dental procedures like tooth extraction, surgery, puncture wounds, or blunt injuries in the affected area. In this manuscript, we report a case of AV malformation involving the left cheek and buccal mucosa region in a 37-year-old male patient who reported uncontrolled bleeding after trauma. This report highlights the management of AV malformation in an emergency by facial artery ligation and surgical excision.

Keywords: Arteriovenous malformations, cheek, Colorado microdissection tips, hemorrhage, vascular diseases

INTRODUCTION

Arteriovenous malformation (AVM) is a rare vascular anomaly involving a complicated network of arteries and veins. It is usually congenital but can present as a consequence of trauma. In 1982, Mulliken and Glowacki[1] categorized vascular lesions into two types: hemangioma and vascular malformations. Rapid cellular propagation and endothelial hyperplasia are critical, distinctive features of hemangiomas. These structures are absent during birth and undergo rapid growth during the first year of life. Endothelial cells in AVM have a standard rate of replacement and are congenital, which are apparent later in life. The malformations experience rapid enlargement due to trauma or hormonal fluctuations. The increase in the size of these lesions is caused by alterations in pressure and flow, extension of vascular channels, redirection of blood flow, and the growth of collateral blood vessels, rather than cellular proliferation.[2] Based on blood turbulence, vascular malformations are categorized into low- and high-flow lesions. The low-flow category includes venous malformation, capillary, and lymphocytic lesions while high-flow encloses arterial and veno-arterial malformation.[3]

AVMs form 1.5% of all endothelial anomalies, of which 50% are seen in the oro-maxillary facial region.[4] AVM causes functional and cosmetic issues like asymmetry, pain, bone destruction, and unanticipated hemorrhage. AVMs are characterized histologically by abnormal arteriovenous shunts without typical interconnecting capillary beds. These vasculatures have numerous arterial and venous compartments, endothelial proliferation, and giant cells.[5]

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Received: 06 September 2023, **Revised:** 26 March 2024, **Accepted:** 28 March 2024, **Published:** 24 July 2024

Access this article online	
Website: www.njms.in	Quick Response Code
DOI: 10.4103/njms.njms_159_23	

How to cite this article: Mahajan A, Nagpal T, Shah A, Daithankar S. Excision of arteriovenous malformation in an emergency situation - A case report. Natl J Maxillofac Surg 2024;15:319-22.

This case report describes the emergency management of an AV malformation in a 37-year-old male patient triggered by trauma.

CASE REPORT

A 37 year male patient reported to the department of oral and maxillofacial surgery with a complaint of continuous bleeding from the left buccal mucosa due to a history of trauma a day prior. The patient explained about the presence of swelling over the left buccal mucosa for ten years, which gradually increased in size. On examination, extra-oral swelling measured approximately 3 cm × 4 cm, extending supero-inferiorly from the malar process to the lower border of the mandible and anteroposteriorly from the corner of the mouth to the angle of the mandible [Figure 1]. The swelling was soft, compressible, fluctuant, and pulsatile with bruit audible on auscultation.

Ultrasonography(USG) Doppler imaging [Figure 2] demonstrated a large multiloculated cystic lesion measuring about 4.3 * 2.6 cm with intense vascularity and multiple dilated tortuous channels. The feeding vessel was assumed to be a branch of the facial artery seen at the cranial aspect of the lesion. Despite the application of a pressure pack, the bleeding remained uncontrollable.

Figure 1: Extraoral swelling over the left cheek region

After obtaining consent patient was taken for excision of the lesion under general anesthesia. An incision was placed with a Colorado microdissection tip (N103A) over the left border of the mandible, and the subplatysmal flap was reflected. The extent of the lesion was extending upwards over the masseter muscle. The facial artery, vein, and retromandibular vein were identified and ligated. The extraoral incision was placed below the zygoma. The lesion was seen infiltrating the masseter muscle, which was excised [Figure 3] along with the masseter muscle and sent for histopathology. The flap was repositioned, and hemostasis was achieved [Figure 4]. Histopathological sections demonstrated dilated, congested vessels surrounded by fibro-connective tissue and lymphocytic infiltration with areas of hemorrhage. [Figure 5]. Features suggested AVM. The swelling reduced postoperatively [Figure 6].

DISCUSSION

AVMs are fast-flowing abnormalities resulting from the redirection of blood between the arterial and venous systems, mainly due to abnormal capillary beds.

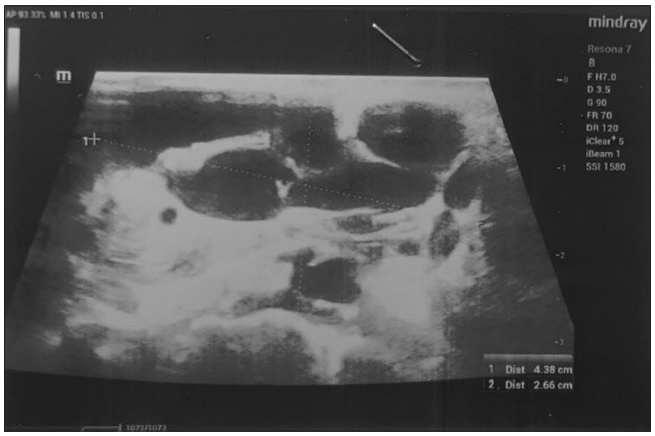

Figure 2: USG image of the left cheek region

Figure 3: Intraoperative picture demonstrating the excision of mass in toto

Figure 4: Closure of skin with Vicryl and Prolene

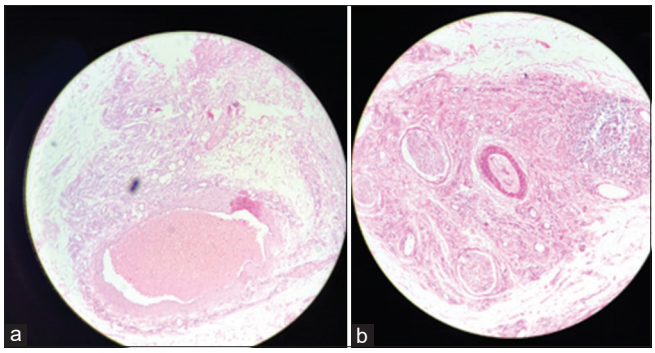

Figure 5: (a) The H and E-stained section shows large dilated blood vessels with RBCs and the vessel is surrounded by fibro-connective tissue (10 × magnification). (b) A H and E-stained section shows the presence of capillaries, veins, and nerve bundles surrounded by fibro-cellular connective tissue and lymphocytic infiltration (10 × magnification)

Holt *et al*.[6] suggest that AVMs arise from various types of trauma. The expansion is due to alterations in pressure, ectasia, shunting, and collateral proliferation. In this case, bleeding was initiated by a traumatic bite. Diagnostic techniques like computed tomography (CT), magnetic resonance imaging (MRI), and resonance angiography can be used for these vascular lesions. AVMs are identifiable as highly enhancing lesions on a CT scan when iodinated contrast is used, revealing enlarged feeding and drainage vessels. MRI is effective independently and is also a prelude to angiography. High- and low-flow lesions are precisely depicted with the help of MRI sequences. High-flow lesions in T1 and T2 sequences display signal flow gaps resembling serpentine patterns. In our case, the patient underwent color Doppler USG, showing high flow in the facial artery.[7,8] Management of AVM is challenging due to the propensity of deceased vessels to replace healthy tissue and the high flow rate. The treatment options for AVM include surgery, vascular embolization, or a combination. Wide resections pose potential risks due to the significant intraoperative blood loss. The objective of embolization is to obstruct the feeder vessels using materials like polyvinyl alcohol, muscles, gel foam, cyanoacrylate, and collagen.[9,10] In our case, intraoperative bleeding was not much as a Colorado microdissection tip was used which provides safe and meticulous dissection with adequate hemostasis. Also, it has the advantages of less tissue necrosis and less postoperative complications.[11,12] We did not use sclerosing agents, as it was an emergency situation due to active intraoral bleeding.

CONCLUSION

Fifty percent of AVM occurs in the maxillofacial region. AVMs are usually wrongly diagnosed, which causes oral bleeding leading to fatal complications. All investigations need to be performed before any oral procedure. Imaging and histopathology help to precisely characterize the lesion, which aids in the evaluation and prognosis and deciding a suitable treatment strategy. The integration of medical and surgical procedures poses challenges, necessitating a multi-departmental approach.

Figure 6: Postoperative view of the patient during follow-up

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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