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Case Report

Surgery Section

Surgical Management of a High Flow Auricular Arteriovenous Malformation Presenting with Life-Threatening Haemorrhage: A Case Report

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ABSTRACT

High-flow Arteriovenous Malformations (AVM) of the auricular region is rare but difficult-to-treat vascular anomalies. As a benign entity, management of the lesion should balance aesthetic considerations, long-term function, and the avoidance of life-threatening complications. A 35-year-old male presented to the emergency department in shock with pulsatile bleeding from the left ear, following a history of progressive swelling and recurrent spontaneous haemorrhages from the left auricular region. On examination, a bruit could be heard, suggesting a high-flow AVM, and the patient was adequately resuscitated with blood products. Magnetic Resonance Angiography (MR Angiography) was suggestive of an enlarged external carotid artery with an extensive AVM around the left auricular and temporal region. A multidisciplinary team comprising plastic surgeons, interventional radiologists, surgical oncologists, and vascular surgeons reviewed the case and decided on surgical excision as the best course of action. Surgical extirpation was successfully performed along with external carotid artery ligation to control the haemorrhage with the defect being reconstructed using split thickness grafts. The patient has been on follow-up for two months with no evidence of disease recurrence. Rare locations of high-flow AVMs should always be discussed with a multidisciplinary team to determine the best course of action among the available options for the patient.

Keywords: Excision, External carotid artery, Multidisciplinary team, Split thickness graft, Temporal region

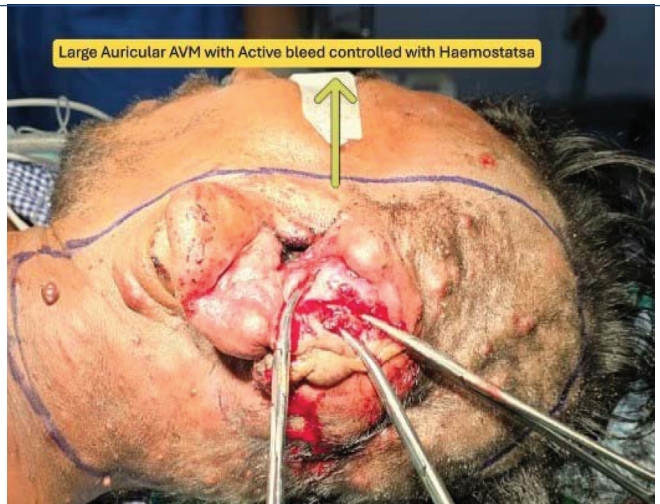
CASE REPORT

A 35-year-old male presented to the emergency department with a swelling in the left periauricular region associated with recurrent episodes of bleeding. His vital signs were as follows: Blood Pressure (BP) 80/40 mmHg, Pulse Rate (PR) 120 bpm, and Haemoglobin (Hb) 3 g/dL.

The patient reported a swelling in the left periauricular region that had been present since birth and had gradually increased in size. It was associated with changes in skin colour and intermittent episodes of spontaneous, painless bleeding. There was no history of trauma or similar swellings elsewhere on the body.

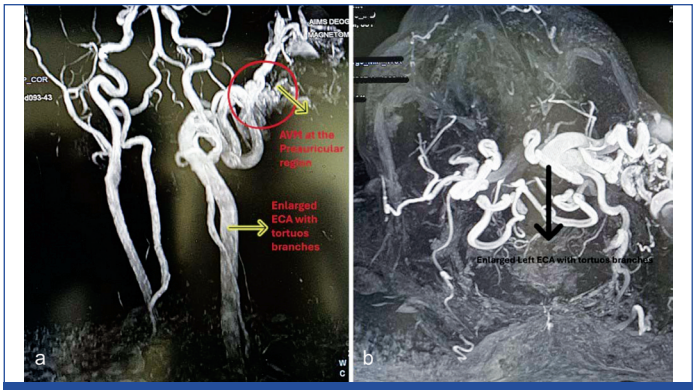
On local examination, a pulsatile swelling measuring 15×6 cm over the left auricular region was seen. The lesion extended superiorly involving the temporoparietal scalp around 8 cm below the vertex, anteriorly around 5 cm from the lateral canthus, posteriorly 6 cm from the external occipital protuberance, and inferiorly till 4 cm from the angle of the mandible. The lesion was grossly involving the external ear. The bleeding was controlled by applying multiple artery forceps [Table/Fig-1]. On palpation, the swellings were soft, non-tender, pulsatile, and compressible. Bleeding was present, and a bruit was heard on auscultation. Skin discolouration was noted, but there were no ulcerations over the swellings. The external auditory canal and tympanic membrane were normal. Based on the patient's history and clinical findings, a diagnosis of high-flow arterial malformation was made. According to Schobinger's classification of AVM, the patient belonged to stage III [1]. The patient was resuscitated with intravenous (i.v.) fluids and transfused with seven units of Packed Red Blood Cells (PRBCs) over three days and MR angiography was planned.

MR angiography revealed a lobulated soft tissue mass showing multiple serpiginous flow voids (bag of worms) in the skin and subcutaneous plane in the left retroauricular region. The nidus showed feeding branches from the ipsilateral external carotid artery [Table/Fig-2a]. A few branches were also seen arising from the superficial temporal artery and occipital artery. The left external jugular vein and retromandibular vein were dilated. In contrast, early filling of enlarged draining extracranial veins was observed. A few non-enhancing regions within were suggestive of thrombus. No intracranial extension was observed [Table/Fig-2b]. Unfortunately, digital subtraction angiography was not available in the institute.



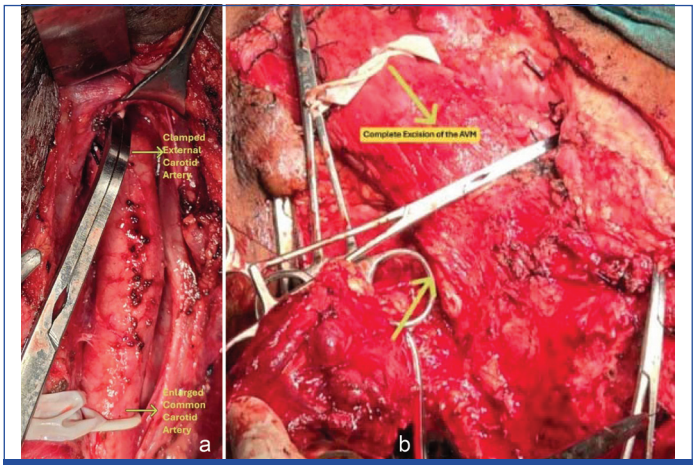
[Table/Fig-1]: Large pulsatile swelling over left auricular and peri auricular region with active bleed controlled with haemostats.

With a plethora of management options, which include ethanol embolisation, sclerotherapy, intravascular clips, and surgery, an interdisciplinary meeting was held with a team of plastic surgeons, radiologists, surgical oncologists, and vascular surgeons. On discussion of the imaging findings, the large feeding vessel, and active bleeding complication, patient was planned for surgical excision.



[Table/Fig-2]: a) MR Angiography coronal view showing enlarged and hypertrophied common carotid and external carotid artery with multiple enlarged branches in the left auricular region; b) Axial view of the angiography showing clusters of arterioles and capillaries arising from the external carotid artery.

The incision was designed elliptically, centred around the left external ear, to include the whole of the lesion. The neck portion of the incision was utilised to gain access to the left external carotid artery. Intraoperatively, it was observed that the left external carotid artery was significantly dilated, equivalent in size to the common carotid artery. The procedure began with ligation of the left external carotid artery to gain vascular control. This step was only undertaken to control the life-threatening haemorrhage and reduce the blood flow to the malformation during excision as a last resort. The entire external ear, along with the cartilage, was excised, and the AVM tissue in the ear, neck, and temporal region of the scalp was removed [Table/Fig-3a,b]. Facial nerve identification was not required as we were well above the superficial parotid fascia. The marginal mandibular nerve was identified 2 cm below the angle of the mandible and preserved. The neck was reconstructed using local tissue rearrangement, whereas all other defects were covered with split thickness skin grafts. Margins were assessed using frozen section to ensure completeness of resection. Total operative duration was 230 minutes. Total blood loss was around 1800 mL, and the patient spent approximately 24 hours in the ICU for observation only. The patient was discharged on postoperative day 12 uneventfully.

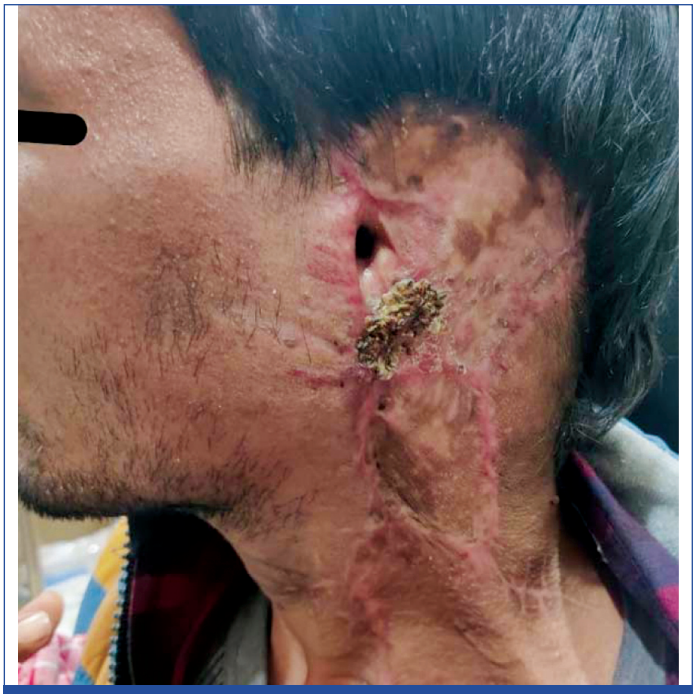


[Table/Fig-3]: a) External carotid artery clamped to decrease supply to feeding vessels and decrease blood loss; b) Complete excision of the arteriovenous malformation with amputation of the external ear.

The patient was followed for two months, during which no recurrence of the AVM was noted [Table/Fig-4]. Postoperative histopathology revealed dilated arteries and veins with haemorrhage and thrombi in areas in between, and an overall impression of a benign vascular lesion, likely an AVM. He was planned for ear reconstruction after three months postoperatively.

DISCUSSION

AVMs are believed to result from errors in vascular development during the 4th to 6th weeks of embryogenesis, due to the failed regression of AV channels in the primitive plexus [2]. High-Flow



[Table/Fig-4]: Follow-up at 2 months showing no evidence of recurrence.

Vascular Malformations (HFVMs) are rare lesions arising from errors in vascular morphogenesis. They are characterised by AV shunting under arterial pressure, leading to haemorrhage, ischaemia, pain, and tissue breakdown [2]. Local ischaemia and trauma are also thought to contribute to their pathogenesis and progression. AVMs lack a capillary bed, causing direct shunting of blood from arteries to veins via a fistula or a nidus, leading to uncontrolled bleeding, tissue destruction, and deformity [3]. While AVMs are present at birth, they often remain asymptomatic until childhood or later and typically worsen over time [3].

A systematic review of 24 studies involving 980 patients reported recurrence rates of 11% for arteriovenous malformations, 5% for venous malformations, and 9% for lymphatic malformations. Importantly, recurrence was significantly higher following subtotal resection than total resection (28% vs. 5%; OR 0.14, p=0.02), highlighting the importance of complete excision whenever anatomically feasible. These findings support the effectiveness of surgical management while emphasising the need to balance complete resection with the risk of complications, particularly in extensive or anatomically complex lesions [4].

Imaging, such as MRI and MR angiography, helps define the lesion's extent, while angiography is reserved for unclear cases or preoperative planning. AVMs appear as tortuous, dilated vessels with AV shunting and enlarged draining veins [5].

A recent systematic review of congenital vascular malformations of the external ear reported that swelling and bruit or thrill were common clinical presentations, with most cases classified as Schobinger stage II. Angiography was the most frequently used diagnostic modality, while ethanol embolisation was the predominant treatment, followed by surgical excision with preoperative embolisation. The reported recurrence rate was 7.4% over a median follow-up of 15 months. These findings highlight the importance of appropriate vascular imaging and individualised treatment planning, with the choice of embolization, surgical excision, or a combination depending on the extent and characteristics of the lesion [6].

Management of AVMs is challenging due to their diffuse nature, involvement of vital structures, and the risk of significant blood loss during treatment. The goal of treatment is not to cure but to control complications like pain, bleeding, ulceration, or disfigurement. Surgical approaches should balance resection with minimising deformity, as extensive surgeries can lead to worse outcomes than the original lesion [7].

CONCLUSION(S)

The HFAVMs of the head and neck are rare and hard to cure and can present with massive haemorrhage and pulsatile masses. Focus should remain on symptom relief, aesthetics, and, if possible, function preservation. Careful planning and long term follow-up are vital given the risk of recurrence and intracranial involvement.

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