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Arteriovenous Malformation of the Externalear: A Case Report

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Abstract **Case Report**

Arteriovenous malformation (AVM) refers to a direct connection between an artery and vein without capillary involvement, predominantly occurring within the intracranial region. However, occurrences of extracranial AVMs, particularly in the external ear, are relatively rare. Recently, we encountered a case involving an AVM in the external ear. An 18-year-old female patient presented with pulsatile persisting for the past 48 months, along with a reddish, pulsatile mass on the left external ear. We successfully managed the AVM by completely excising it and covered it by a thick skin graft following preoperative selective embolization.

Keywords: Arteriovenous malformation – Embolisation – nidus – surgery.

INTRODUCTION

An arteriovenous malformation (AVM) is characterized by a direct connection between an artery and vein without capillary involvement, primarily occurring in the intracranial region, lung, and kidney [1]. However, AVMs outside the cranium are rare, with the most common extracranial sites being the cheek, ear, nose, and forehead [2]. The preferred treatment for AVMs involves preoperative selective embolization followed by complete excision [1, 2]. We recently encountered a case of an AVM in the external ear and provide a brief literature review along with presenting this case.

CASE REPORT

An 18-year-old female patient was assessed at the plastic surgery department for a reddish, pulsatile mass located on the right external ear. The patient reported experiencing pulsatile tinnitus for the past 48 months, along with the presence of the reddish, pulsatile mass on the right external ear. Shedid not have any significant medical history. A reddish and pulsatile mass, measuring 36 mm x 12 mm, was observed at the right helix and antitragus, along with another mass measuring 14 mm x 30 mm at the anterior portion of the right antihelix (Fig 1). Auscultation revealed a bruit over the mass, and the surronding tissue appeared erythematous and edematous.

Magnetic resonance imaging (MRI) revealed abnormal signal voiding intensity of the mass on axial T2-weighted images, with some areas showing high signal intensity extending to the left external ear (Fig 2A). Selective angiography confirmed the diagnosis of arteriovenous malformation (AVM), demonstrating a diffuse network of shunts with dilatation and tortuosity of the feedingarteries and draining veins. The AVM was supplied by two main arteries: the posterior auricular artery and the superficial temporal artery (Fig 2B). Super selective embolization of the posterior auricular artery was performed using Curaspon.

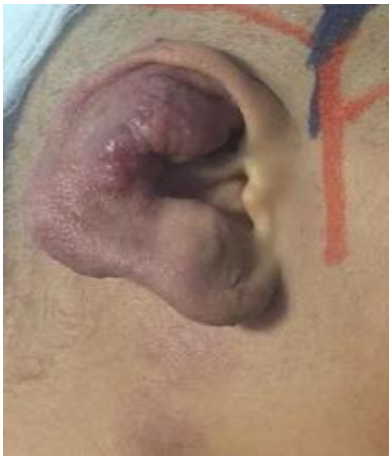

Fig 1: Preoperative photograph of the right external ear

Following control of the inflammatory reaction, the patient underwent excision of the AVM 24 hours later.

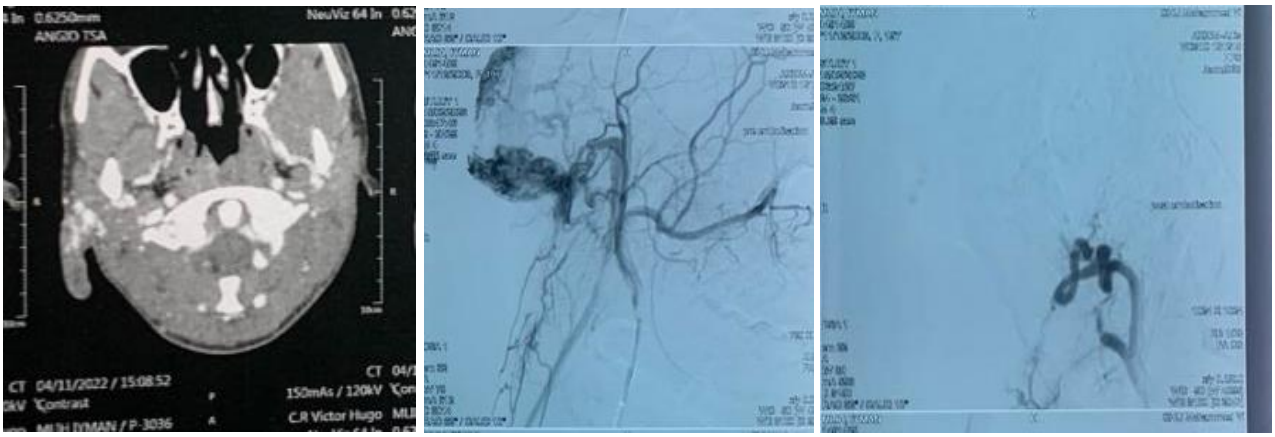

Fig 2: Preoperative findings of magnetic resonance imaging (MRI) and angiography of AVM. The preoperative MRI finding (axial T2-weighted image) shows principally abnormal signal voiding intensity of the mass (arrow) with some high signal intensity at the left external ear (A). The preembolization of angiography shows that the two branches of the superficial temporal artery supply the anterior portion of the helix and one branch of the posterior auricular artery supplies the cymba conchae of the AVM (B). AVM, arteriovenous malformation; ECA, external carotid artery; STA, superficial temporal artery; PA, posterior auricular artery; OA, occipital artery

The excision of the AVM included identification and ligation of the two feeding vessels, followed by a skin graft (Fig 3). Histopathological examination of the surgical specimen revealed multiple dilated arterial and venous channels with thromboses, direct arteriovenous communications, and mixed capillary and cavernous malformations lined withthin-walled channels.

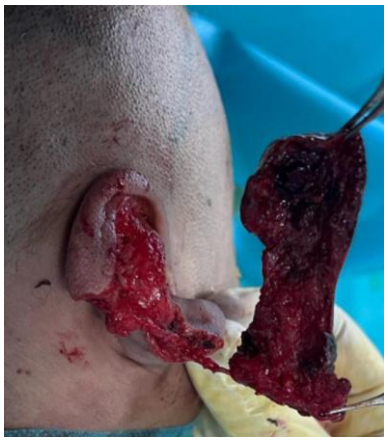

Fig 3: Preoperative photograph showing the feeding artery

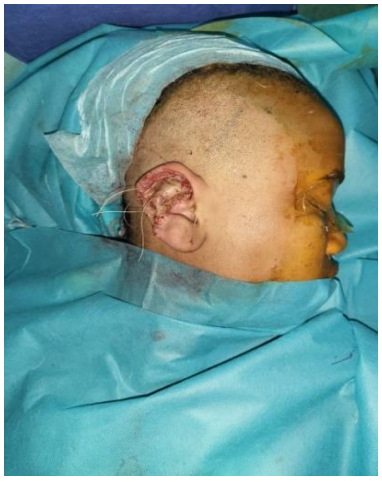

Fig 4: Preoperative photograph showing the final result after proceeding to a thick skin graft

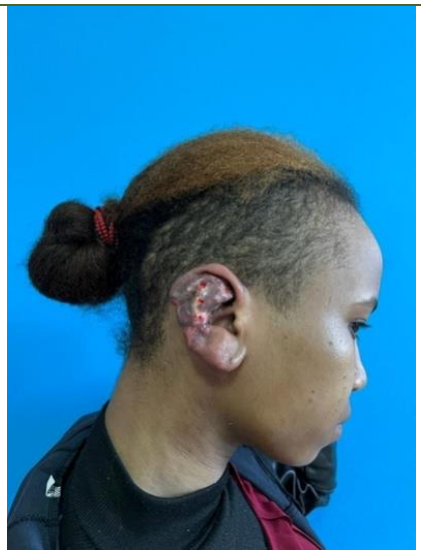

Fig 5: Photograph showing the results after one month of the excision and the skin graft

After a one-year follow-up period, the patient showed no signs of recurrent AVM.

DISCUSSION

An arteriovenous malformation (AVM) arises from the failure of regression of arteriovenous channels in the primitive retiform plexus during the fourth to sixth weeks of gestation. While present at birth, AVMs are typically not clinically significantinitially, aside from cosmetic concerns. The incidence of AVMs has been reported as 1:1.5 (male:female) [1]. Over time, AVMs can enlarge due to increased blood flow and the formation of collateral vessels. Histopathologically, AVMs differ from hemangiomas in that they do not exhibit cellular hyperplasia but rather progressive ectasia of abnormalvessels [3].

Clinical diagnosis of an auricular AVM is often based on history and physical examination findings. AuricularAVMs are classified using the Schobinger staging system, with the present case falling into stage two [1]. Ultrasonography confirms the presence of an AVM [4], while magnetic resonance imaging delineates soft tissue involvement in larger malformations, and computed tomography investigates skeletal involvement [1]. Selective angiography is particularly useful for identifying and embolizing the specific arterial supply of an AVM [3].

Although the management of auricular AVMs had previously been limited to single or few case reports, recent studies by Ferreira *et al*., and Wu *et al*., have provided valuable insights into treatment approaches for auricular AVMs [2, 8]. While small and asymptomatic auricular AVMs may not require treatment, managing large symptomatic auricular AVMs is often challenging.

The optimal treatment for auricular arteriovenous malformations (AVMs) involves a combined approach of selective embolization and complete excision. It is recommended to perform complete excision of the auricular AVM within 48 hours after embolization to minimize collateralization [2, 5, 6]. Other treatment modalities such as laser therapy, steroid administration, or irradiation have not shown effectiveness [1]. Partial excision should be avoided as it is not curative and may lead to recurrence due to revascularization and the development of new collateral circulation [1, 2]. Proximal ligation of arterial vessels aloneis not recommended because it can result in the formation of new collateral circulations. While super selective embolization alone may be used for palliation, it is rarely successful due to the formation of new collateral vessels [1, 2, 5]. In this case, successful treatment of the patient's external ear AVM was achieved through complete excision following preoperative selective embolization.

CONCLUSION

For small, asymptomatic auricular arteriovenous malformations (AVMs), treatment is typically unnecessary. However, for large, symptomatic auricular AVMs, the optimal treatment may involve complete surgical excision following super selective embolization.

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