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Case Report

Recurrent head and neck arteriovenous malformations: A case report ☆

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ABSTRACT

Head and neck arteriovenous malformations are rare, congenital, and high-flow vascular malformations characterized by abnormal communication between feeding arteries and draining veins without intervening capillaries. Arteriovenous malformations are considered the most dangerous type of vascular malformation because progressive symptoms and infiltration can result in potentially life-threatening consequences. Left untreated, arteriovenous malformations can cause significant cosmetic deformities, severe bleeding, and high-output cardiac failure associated with arteriovenous shunting. The effective treatment of arteriovenous malformations located in the head and neck region is quite challenging due to high rates of recurrence and potentially lethal complications. We describe a case presenting with large arteriovenous malformations in the face and neck. Despite attempting several treatments, including external carotid artery ligation and embolization with liquid embolic agents, the patient continued to experience recurrence and symptoms of bleeding and pain. After admission, reconstructive plastic surgery was performed, supplemented by percutaneous direct puncture embolization, using glue injected into the venous and transarterial embolization. The patient was discharged with clinical recovery. Digital subtraction angiography remains the gold standard for assessing symptomatic and aggressive arteriovenous malformations, both before and after treatment. The treatment of head and neck arteriovenous malformations often requires a multidisciplinary approach to achieve the best clinical results.

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Introduction

Head and neck (H&N) arteriovenous malformations (AVMs) are high-flow, congenital, vascular anomalies consisting of a complex system of vessels that result in a direct connection between feeding arteries and draining veins, forming a nidus. AVMs represent only 1.5% of all vascular anomalies [1,2] and are often identified in the H&N region (47.4%) and in the extremities (28.5%) [3]. Signs and symptoms of AVMs include local erythema, hyperthermia with prominent pulsations, a palpable thrill, and an audible bruit. Digital subtraction angiography (DSA) is the current gold standard for defining the morphology, nidus, and supportive vasculature of AVMs because DSA is more accurate and less invasive than other modalities, including conventional angiography, and provides the best spatial and temporal resolution for studying the vascular anatomy [4]. Current AVM management strategies aim to reduce the risk of bleeding and include conventional surgery and endovascular treatment. The complete removal of AVMs is challenging, and the rate of recurrence is high. Embolization, used either alone or together with surgery, can be an effective method for improving symptoms. Embolization is typically recommended to be followed by surgical resection, performed either immediately or within a short time. We present a case with H&N AVM treated with embolization. However, a single treatment is often insufficient to resolve these high-flow lesions due to the recruitment of arteries from surrounding tissue, which can induce the aggressive growth of the remaining nidus and result in worsening disease [5–7].

Case report

A 46-year-old man presented at our hospital with a large, wet, rigid swelling area in the left ear region, characterized by minor bleeding and bruit. Although the patient stated that the swelling was painless, old surgical scars in the left corner of the lower jaw caused discomfort when the patient was lying on the left side (Fig. 1). Clinical examination did not reveal any signs of nerve damage associated with the left corner of the lower jaw. The patient's medical history included the prior treatment of an AVM in the neck area using an external carotid artery ligation approach in 1995 and the diagnosis of recurrent AVM in 2007 (resulting in the removal of the left ear) and 2017. In 2019, the patient underwent embolization at our hospital to prevent recurrence (Fig. 2). In 2021, we performed an emergency embolization procedure due to progressive lesion enlargement, accompanied by severe bleeding in the corner of the left jaw (Fig. 3). After the emergency embolization procedure, no further bleeding occurred. His family history shows no evidence that this represents a hereditary condition. We opted to attempt a third embolization procedure.

DSA revealed an AVM in the scalp behind the left ear consisting of many branches originating from the left external carotid artery (tied) that formed anastomoses with branches originating from the thyrocervical trunk artery and opposite carotid artery. The AVM was classified as a Yakes type IV, which is defined as a network of innumerable arteriovenous shunts without a defined nidus infiltrating tissues, with capillary beds interspersed among the innumerable shunts. The left subclavian artery was accessed using a co-axial system consisting of a Chaperon 6F (Microvention) sheath and a diagnostic 5F vertebral catheter (Merit Medical) and 0.035″ wire (Terumo) to generate a roadmap. A 1.5F Magic microcatheters (Balt) and 0.007″ hybrid microwire (Balt) were used to selectively approach the branches of the left thyrocervical trunk artery. Embolization was performed 3 times using a combination of Histoacryl glue (B.Braun) and Lipiodol Ultra-Fluid (Guerbet) (Figs. 1-4). This mixture was also injected percutaneously directly into the AVM venous pouch and nidus while the ipsilateral jugular vein was compressed. The final arteriogram revealed the near-complete disappearance of the AVM. Two surgeons removed the necrotic tissue from the left ear area to ensure hemostasis. The patient was followed for 7 days and 7-month follow-up with dry skin tissues (Fig. 5).

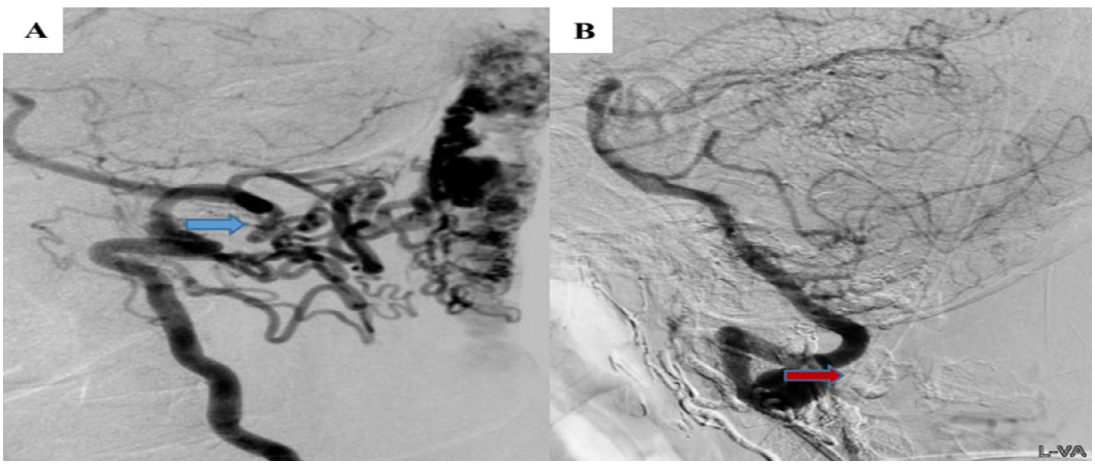

Fig. 1 – Arteriovenous shunts from the left vertebral artery (blue arrow, A) were occluded by glue (red arrow, B) during the initial embolization procedure.

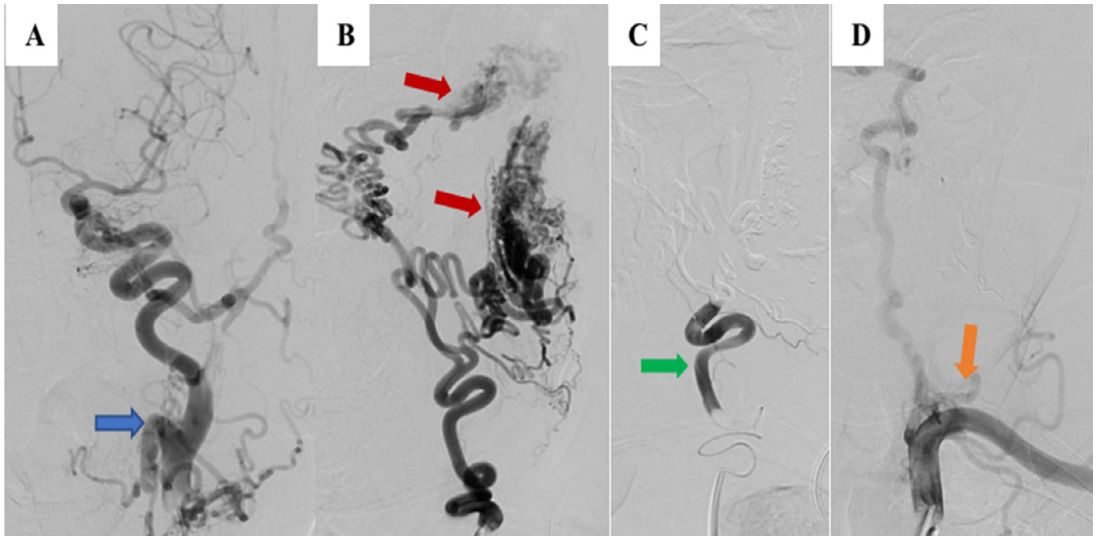

Fig. 2 – Ligation of the left external carotid artery (blue arrow, A). Branches of the thyrocervical trunk artery drained to branches of the left external jugular vein (red arrows, B). Glue was inserted into the thyrocervical trunk artery (green arrow, C). The thyrocervical trunk artery was completely occluded (orange arrow, D) after the second embolization procedure.

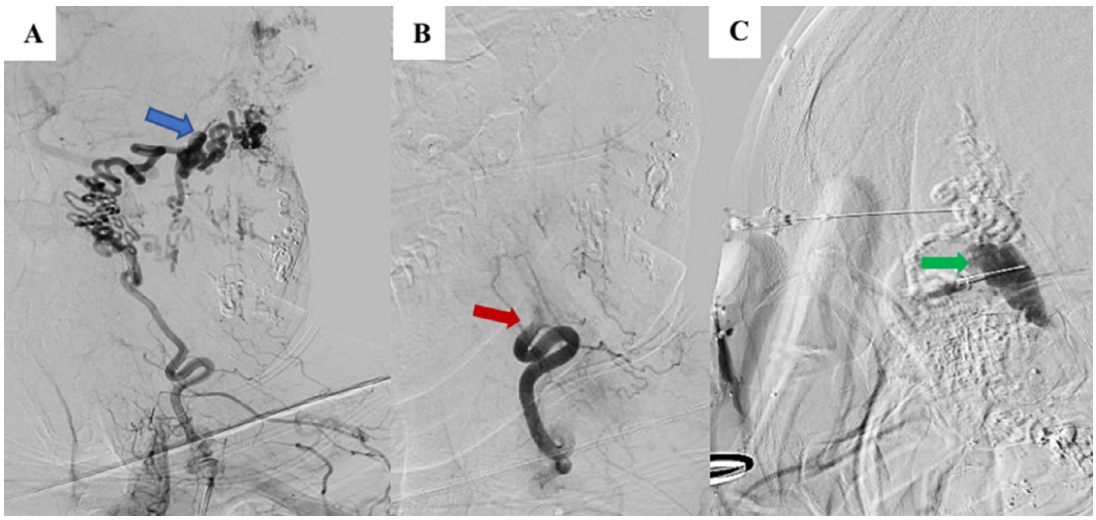

Fig. 3 – New recurrent branches from the thyrocervical trunk artery draining to branches of the left external jugular vein (blue arrow, A). The branches of the thyrocervical trunk artery were occluded (red arrow, B). Percutaneous direct puncture embolization was performed to inject glue directly into the venous pouch (green arrow, C) during the third embolization procedure.

Discussion

AVMs are classified as congenital high-flow vascular anomalies and constitute only 4.7% of all vascular lesions. The central nervous system is the most common location for AVMs, whereas extracranial AVMs are rare [8,9]. AVMs are diagnosed according to history and clinical examination, combined with imaging characteristics. Arteriovenous shunting may have significant hemodynamic impacts, leading to reduced capillary oxygenation, ischemia, local hypervascularity, increased venous pressure, and vascular steal phenomenon. These hemodynamic changes can result in the ulceration or deformation of both soft and bony tissues or the development of congestive heart failure in the absence of lesion control. The Yakes AVM classification system is used in our hospital to evaluate AVM features and classify AVMs into one of 4 types to determine appropriate treatment options.

A retrospective report by Allen showed that the control of complication risk was prioritized in extracranial AVM management over complete obliteration [10]. However, recurrence occurs in greater than 80% of patients who undergo embolization or resection. Recurrent H&N AVMs may occur due because the low pressure of the AVM nidus following incomplete em-

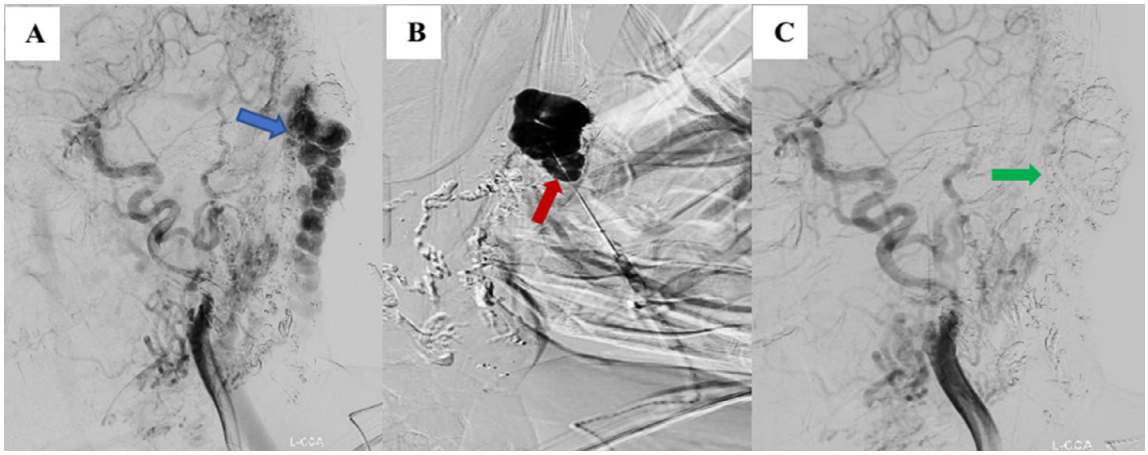

Fig. 4 – Venous drainage from the branches of the left internal carotid artery and the remaining branches of the left external carotid artery (blue arrow, A). Glue was injected into the AVM nidus by direct percutaneous puncture (red arrow, B). The nidus and venous drainage were occluded (green arrow, C) following the third embolization procedure.

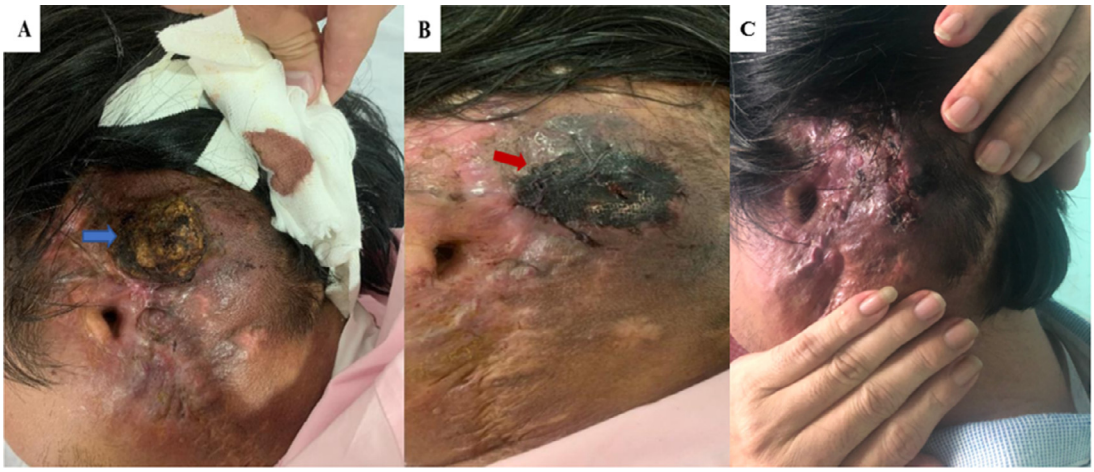

Fig. 5 – Necrotic tissue before (A); after the multidisciplinary approach used during the third embolization procedure (B); and dry skin tissues of the left ear at 7-month follow-up (C).

bolization or resection attracts blood flow from other arteries and promotes the aggressive growth of new arteries in the remaining nidus, which can increase by 50% within the first 5 years [11,12]. Therefore, post-treatment follow-up is essential to detect recurrence in a timely manner.

An interdisciplinary approach is crucial for the successful treatment of complicated H&N AVMs, and vascular embolization plays an important role. In the cases of H&N AVMs referred to our hospital, patients have often reported no close follow-up or continuous embolization, in contrast with our recommendations for long-term AVM management. The lack of follow-up or continuous management allows for the recruitment of reconstituted arterial flow into the nidus, leading to recurrent symptoms that are often more serious than the initial symptoms. Although the ligation of the external carotid artery improved symptoms, this procedure introduced difficulties for both super selective catheterization and direct nidus puncture when attempting to address relapse. The application of embolic agents in the management of AVMs depends on several factors: the vascular characteristics requiring treatment, the Yakes AVM classification, the experience of the treatment team, the availability of embolic agents, and the goals of the procedure [13].

Conclusion

We describe a case with recurrent H&N AVM, despite multiple treatment attempts. We recommend the application of a multidisciplinary approach that includes embolization that follows a detailed plan to prevent the growth and development of complications when treating symptomatic AVMs. Embolization (transarterial or percutaneous) plays an essential role in the successful treatment of recurrent H&N AVMs.

Statement of ethics

Ethical approval was not necessary for the preparation of this article.

Data availability statement

All data generated or analyzed during this study are included in this article and/or its online supplementary material files. Further enquiries can be directed to the corresponding author.

Authors' contribution

Tran Chi Cuong and Le Minh Thang contributed equally to this article as co-first authors. Le Minh Thang, Luu Vinh Qui, Tran-Thi Thanh Tha, and Nguyen Minh Duc contributed to write original draft. Tran Chi Cuong, Nguyen Luu Giang, and Nguyen-Dao Nhat Huy contributed to undergo embolization procedure, collect and interpret the imaging. Nguyen Anh Trung and Tran-Van Lam contributed to perform surgical excision after embolization. Luu Vinh Qui, Tran-Thi Thanh Tha, Duong Hoang Linh, and Nguyen Minh Duc made substantial contributions to collect patient data and clinical data analysis. All authors have read, revised, and approved the final published version of the manuscript. All authors were responsible for submission of our study for publication.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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