

Embolization of Bleeding Pinna Arteriovenous Malformation after Ligation of the External Carotid Artery: A Therapeutic Challenge

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J Clin Interv Radiol ISVIR 2017;1:184–189.

Abstract

A 25-year-old patient presented with bleeding of right pinna arteriovenous malformation (AVM). There was history of ipsilateral external carotid artery (ECA) ligation 10 years back. Subsequent investigations (ultrasound, magnetic resonance imaging, digital subtraction angiography) showed recruitment of complex collaterals from the ipsilateral subclavian artery and vertebral artery feeding the recurrent nidus. The patient underwent two sessions of endovascular embolization and one session of percutaneous embolization. We wish to highlight the feasibility of antegrade embolization in such cases via collaterals and role of direct percutaneous treatment.

Keywords

- ▶ arteriovenous malformation
- ▶ external carotid artery ligation
- ▶ head and neck

Introduction

High-flow arteriovenous malformations (AVMs) develop secondary to a defect in vascular morphogenesis.¹ AVMs in the head and neck region commonly involve the face, while ear is rarely involved.² Small and asymptomatic AVMs generally do not require treatment; however, large symptomatic AVMs require embolization and/or surgery.² Proximal ligation of the vessels without resecting the nidus is catastrophic because it precludes future endovascular access while the nidus continues proliferating due to collateralization.³ We report a case of recurrent large pinna AVM that posed as a therapeutic challenge owing to prior ligation of the external carotid artery (ECA).

Case Report

A 25-year-old woman presented to our hospital with history of swelling over right pinna for the last 10 years and two episodes of bleeding. The recent episode was torrential bleeding requiring prolonged compression for control. There was history of similar bleeding episode 8 years

back for which right ECA ligation was done. Her present ultrasound examination revealed presence of multiple anechoic channels in the right pinna (▶Fig. 1), with contiguous extension into the retroauricular region. Doppler examination revealed color filling in these areas with low-resistance arterial flow, consistent with AVM (▶Fig. 1). Some of the channels were extending into the parotid gland. Magnetic resonance examination revealed similar findings in form of heterogenous soft tissue thickening with presence of multiple flow voids in the pinna and periauricular region (▶Fig. 2).

Digital subtraction angiography (DSA) was performed with selective catheterization of the ipsilateral (right) common carotid artery (CCA), internal carotid artery (ICA), vertebral artery, and right subclavian artery (in view of the previous history of ECA ligation). A large AVM nidus was seen in the region of right pinna, which was fed primarily by an enlarged tortuous feeder from right subclavian artery and another feeder from right vertebral artery (▶Fig. 3). Distal part of vertebral artery was not seen opacified, which could be explained by the steal phenomenon due to blood flow diversion toward the AVM nidus. The subclavian artery

received
June 21, 2017
accepted after revision
September 8, 2017
published online
December 14, 2017

DOI <https://doi.org/10.1055/s-0037-1609044>.
ISSN 2457-0214.

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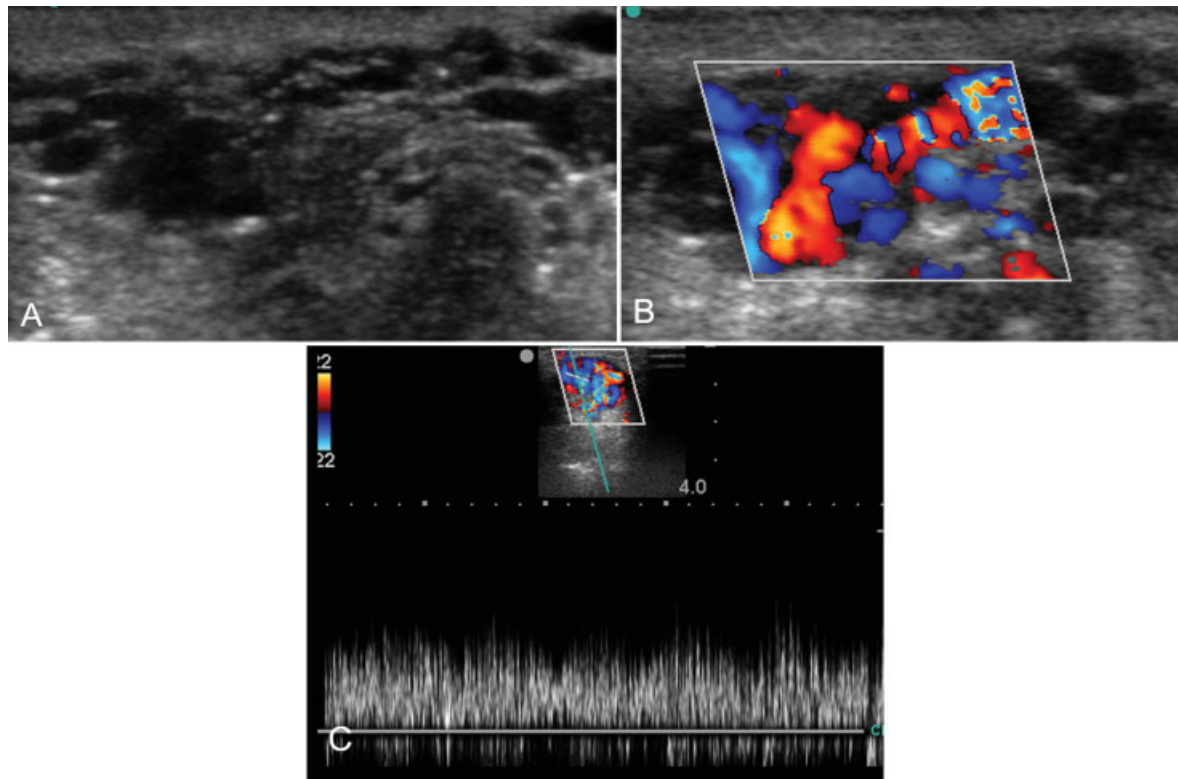


Fig. 1 (A–C) Ultrasound image revealing multiple anechoic channels over the right pinna with variegated color filling on Doppler and bizarre spectral waveform.

feeder followed the expected course of its costocervical branch. Minimal supply was also noted from the branch of ipsilateral ICA. Angiogram of contralateral ECA did not reveal any collateral supply. The prominent tortuous costocervical branch of the right subclavian artery was selectively cannulated and Progret (Terumo Medical Corp.) microcatheter (2.7F, Terumo) was advanced until the distal most optimum point near the nidus. It was embolized using 25% glue in mixture of Lipiodol (0.25 cc *n*-butyl cyanoacrylate, Endocryl, [Baxter] + 0.75 cc Lipiodol [Guerbet]). Post-procedure run was satisfactory revealing significant obliteration of nidus opacification (►Fig. 3). Immediate postprocedure, the patient had an uneventful course and was discharged after 24 hours of observation. To further reduce the size of nidus, one attempt of ultrasound-guided percutaneous embolization with 50% glue was done after about 2 weeks of the first embolization session (►Fig. 4). The procedure was uneventful. However, after 3 days of percutaneous embolization (i.e., after almost 20 days of the first embolization session), the patient reported in emergency with torrential bleeding from pretragus point. The bleeding point was different from the puncture site of percutaneous embolization. Bleeding required prolonged compression and surgical ligation of bleeding punctum for control. The patient was taken for repeat embolization session. Selective check angiogram revealed large tortuous recanalized cervicothoracic feeder of the subclavian artery supplying the AVM nidus (►Fig. 5). There was another less

prominent nearby feeder from the subclavian artery, which on selective cannulation showed reformation of distal ECA with insignificant contribution to AVM nidus. The flow in distal part of the vertebral artery was found to be restored (►Fig. 5F) secondary to successful partial embolization (first session) and subsequent reduction in size of common AVM nidus. This time also, the prominent recanalized costocervical feeder was embolized in the similar way to the first session. The other subclavian feeder to AVM was not embolized due to its insignificant contribution to AVM nidus and anticipated nontarget embolization through reformed ECA. Immediate post procedure angiogram revealed the successful reduction in the opacification of AVM nidus (►Fig. 5). No periprocedural or immediate postprocedural complications occurred. The patient was kept on follow-up. Her follow-up ultrasonography (USG) after 1 and 4 months revealed significant reduction in the AVM nidus with presence of progressive thrombotic obliteration of channels (►Fig. 6). There has been no recurrence of bleeding up to 1 year post last embolization.

Discussion

Arteriovenous malformations are rare in the auricular region.⁴ These are composed of a central nidus with anomalous arteriovenous shunts and fed by a network of collateral vascular channels.⁴ AVM size can increase due to various factors, including trauma, infection, or hormonal influences

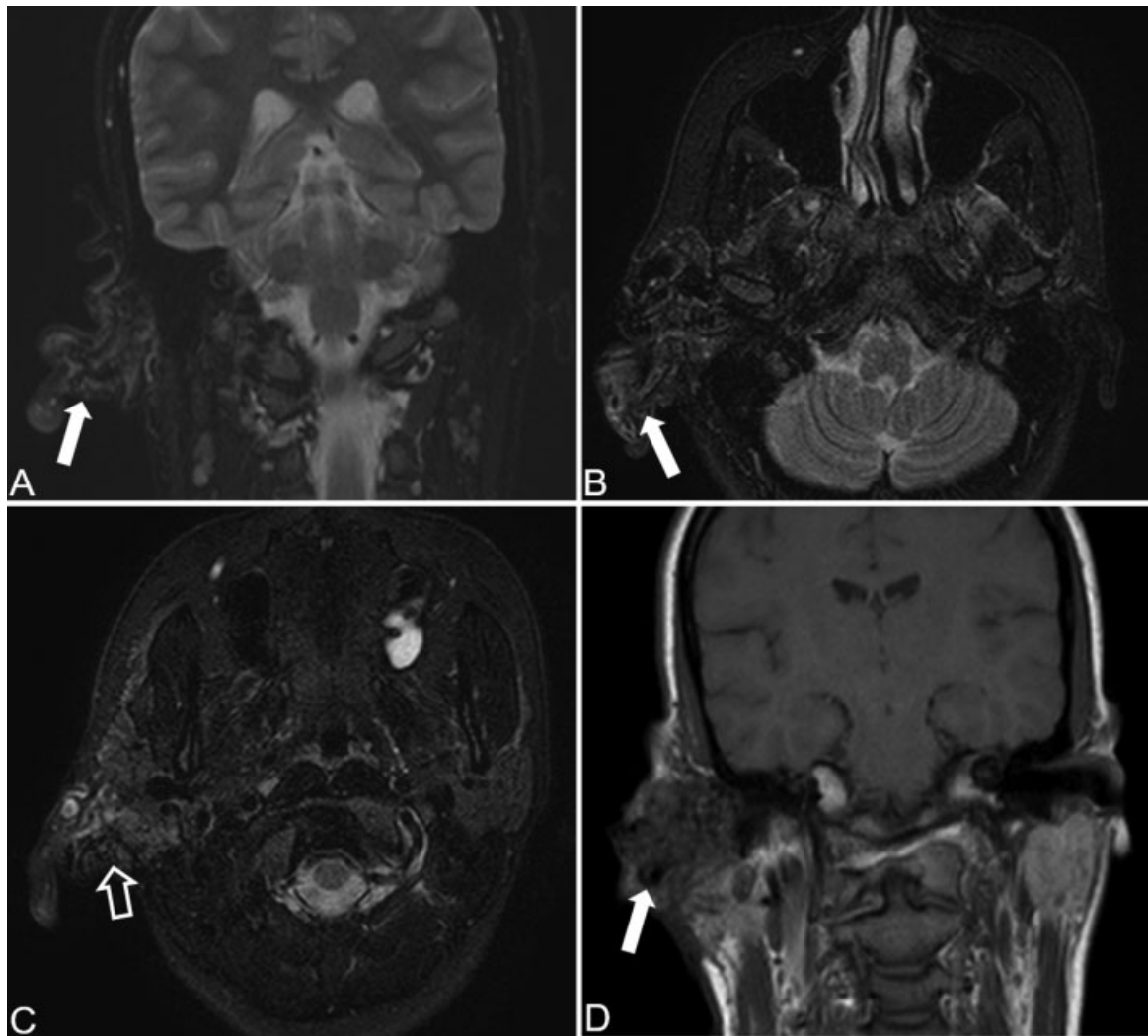


Fig. 2 (A–D) Magnetic resonance imaging STIR coronal (A), axial (B, C), and T1W coronal (D) images reveal heterogeneously bulky right pinna (white solid arrows in A, B, and D) with multiple flow voids and extending into the parotid gland (hollow arrow in image C).

such as puberty and pregnancy.⁵ Presenting symptom may be deformity, pain, ulceration, and bleeding that can sometimes be fatal. The patient first presented with external bleeding from right pinna swelling at the age of 17 years. History and physical examination can provide information that is useful for diagnosis of superficial AVMs of the head and neck. Plain radiograph and computed tomography have limited role as diagnostic tools. Diagnosis is usually made with color Doppler imaging, and magnetic resonance imaging (MRI) is the investigation of choice that helps differentiate the high- versus low-flow malformation as well as to document the extent of lesion.^{1,5} Small and asymptomatic AVMs generally do not require treatment; however, for large symptomatic AVMs, embolization followed by surgery is the treatment of choice.² As per the morphology on angiogram, AVMs can be classified into four types.⁶ Type I refers to arteriovenous fistula morphology with maximum three separate arteries and single draining vein. Type II is arteriovenous fistula type. Type III AVMs have a nidus between the arteries and veins. Type IIIa has a nondilated nidus (seen

as blush or fine striations) and IIIb has dilated nidus. This case conformed to type IIIb on DSA.

Surgical ligation of the proximal vessel, as one of the treatment options of AVM, is obsolete now, as it not only aggravates the lesion by formation of collateral circulation but also precludes the endovascular embolization on follow-up.⁷ ECA ligation used to be the only surgical approach when the patient presented in emergency (with bleeding, airway obstruction or heart failure), but now it has been abandoned.⁸ Though there is immediate relief of symptoms, the long-term results are poor due to development of extensive collaterals, making further management difficult.⁸ In this patient, history of ligation of right ECA was present, which resulted in subsequent progressive increase in the size of swelling due to extensive collateralization of AVM nidus. Even though currently ligation of the feeding artery is seldom done, some patients may have undergone ligation in the past and present with recurrence. Aim of treatment in AVM is to obliterate the nidus, which is usually achieved using liquid embolic agents having endothelial-cidal properties (ethanol

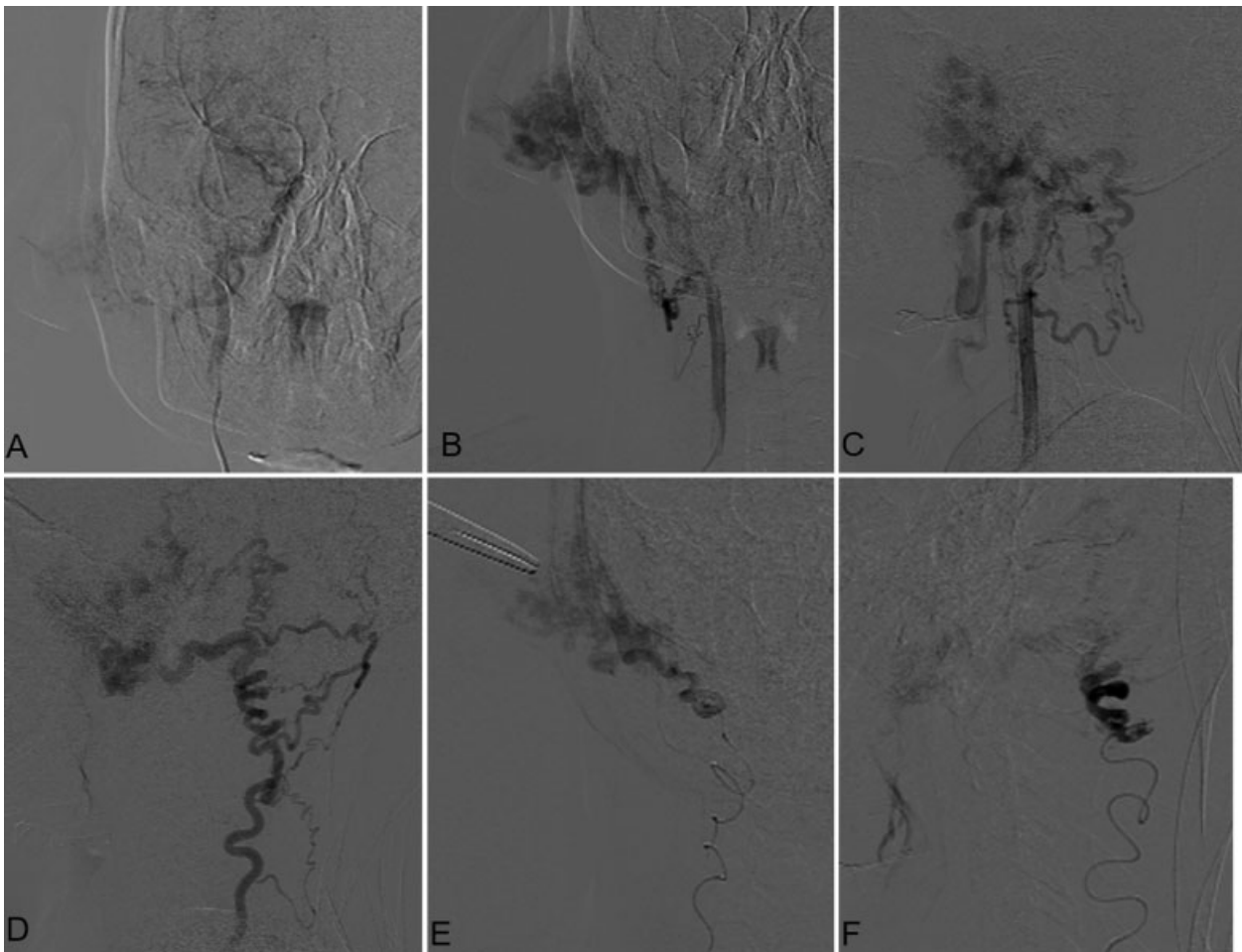


Fig. 3 (A–F) Digital subtraction angiography images reveal feeders from right internal carotid artery (A frontal projection), right vertebral artery (B frontal and C lateral projection), and right subclavian artery (D lateral and E frontal projection) likely through prominent costocervical branch. No blush was seen in the posterior head fossa in the vertebral artery run because of prominent flow toward AVM. The SCA branch was superselectively catheterized and postembolization run (F lateral projection) shows significant reduction in AVM opacification.

and glue).⁹ If the feeder arteries are blocked using coils, this would serve to reduce the flow and short-term symptom relief, but complicate further management similar to what ligating the feeder arteries does.

Collateral blood supply to the AVM nidus after ECA ligation can come from the branches of the internal carotid artery, vertebral artery, and thyrocervical trunk¹⁰ or from the contralateral ECA. In this patient, collateral blood supply

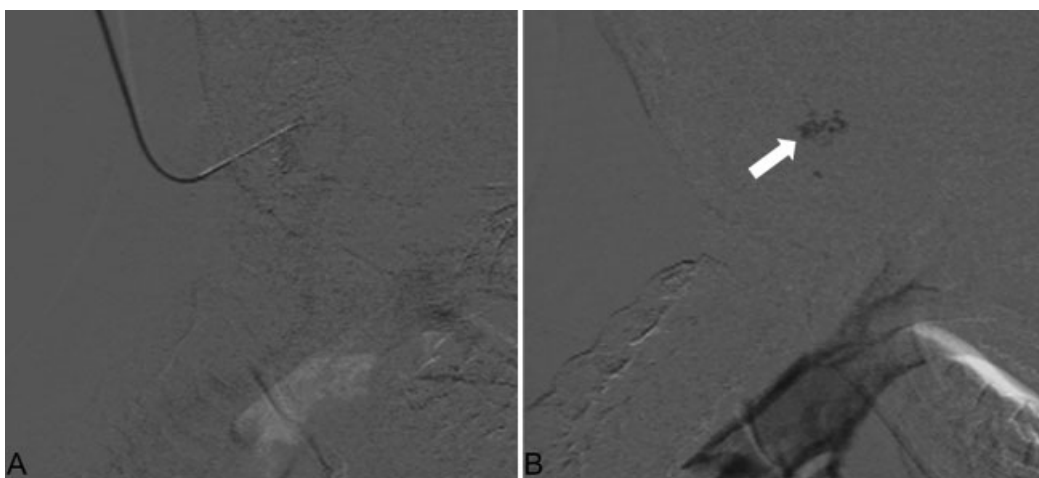


Fig. 4 (A, B) Percutaneous glue (50%) embolization of AVM (arrow showing glue cast).

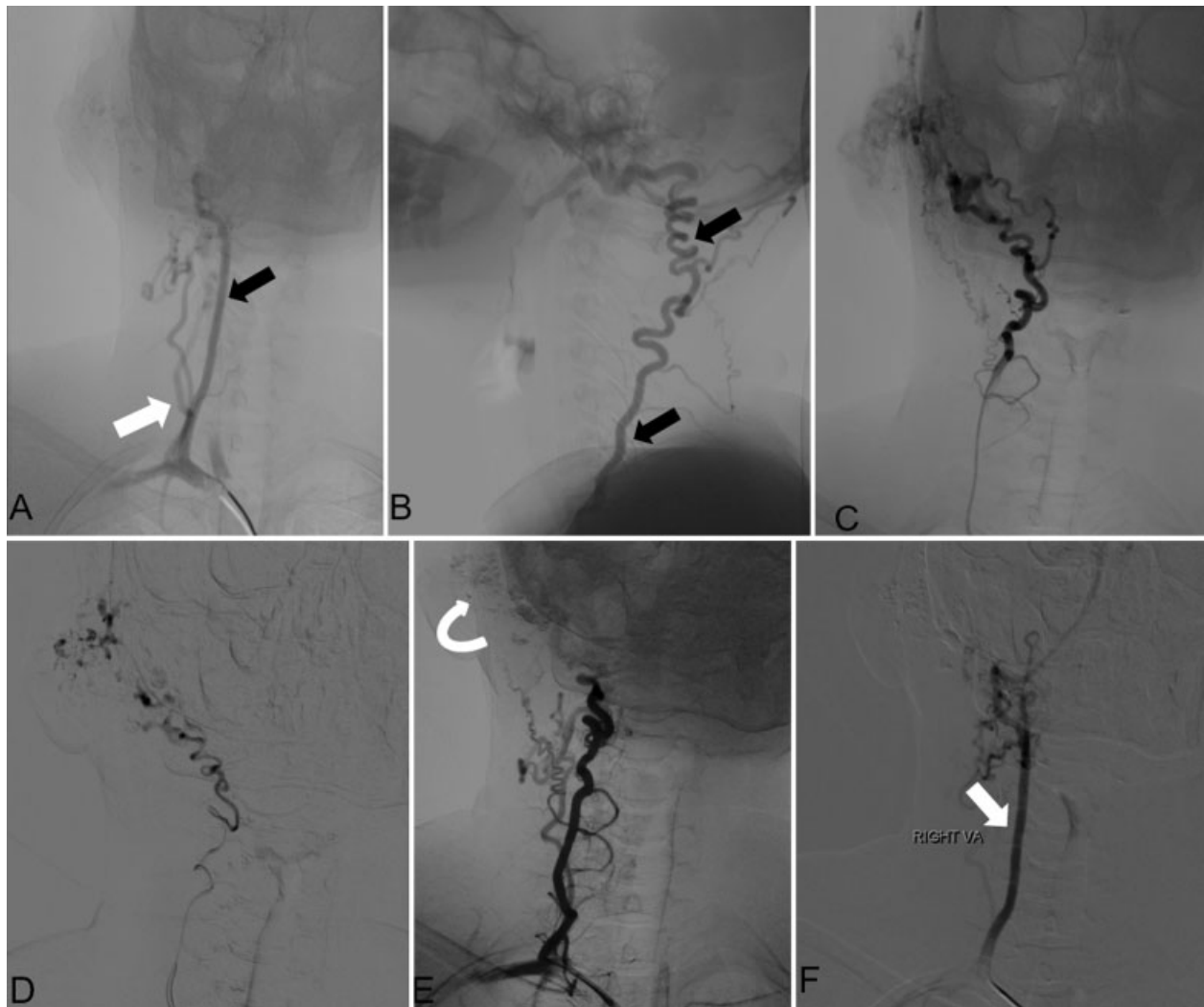


Fig. 5 (A–F) Second session of embolization. Digital subtraction angiography revealing two feeders from right subclavian artery (one of which is recanalized costocervical feeder—black arrows in A and B). After achieving distal most optimum access with Progreat microcatheter and 25% glue embolization (C and D), contrast injection reveals marked obliteration of AVM nidus along with glue cast (curved arrow in E) over pinna. Vertebral artery run (arrow in F) shows restoration of its distal flow probably due to obliteration of the common nidus after first embolization session.

was from the cervical branch of ipsilateral costocervical trunk of the subclavian artery and vertebral artery. There was also absence of contrast filling of vertebral artery distal to collateral circulation on angiogram, which was likely due to steal phenomenon and diversion of blood through collateral channels to AVM nidus. This was restored after successful embolization.

After ECA ligation, endovascular treatment of AVM is challenging due to difficulty in access of nidus, but few innovative approaches have been described in such a scenario. Retrograde puncture of the superficial temporal artery to gain access of AVM nidus is possible if the antegrade approach is not feasible because of prior ECA ligation. Wang et al reported successful retrograde embolization of a bleeding maxillary AVM via the superficial temporal artery after a previous ECA ligation in a 15-year-old boy.¹¹ Retrograde transvenous ethanol embolization is another effective therapy for AVMs, which can be resorted after conventional

treatment options have failed. However, it is associated with high complication rate and can be attempted as the last resort.¹² Surgical reconstruction of the previously ligated ECA is also another feasible option; however, it is associated with inherent risks of immediate AVM engorgement and cardiac decompensation due to sudden rise in cardiac load.⁸ In this patient, good accessible collateral network from the subclavian artery made us capable of approaching the nidus via antegrade route (through catheterization of collateral vessel) followed by successful embolization. Nonrecurrence of bleeding with good reduction in nidus size also exemplifies the role of percutaneous embolization that in this case presumably halted the recruitment of collateral channels and recanalization of feeders after second embolization. As there was no significant supply from the ICA or ophthalmic artery to the AVM, we did not encounter any neurologic complication during or after embolization.

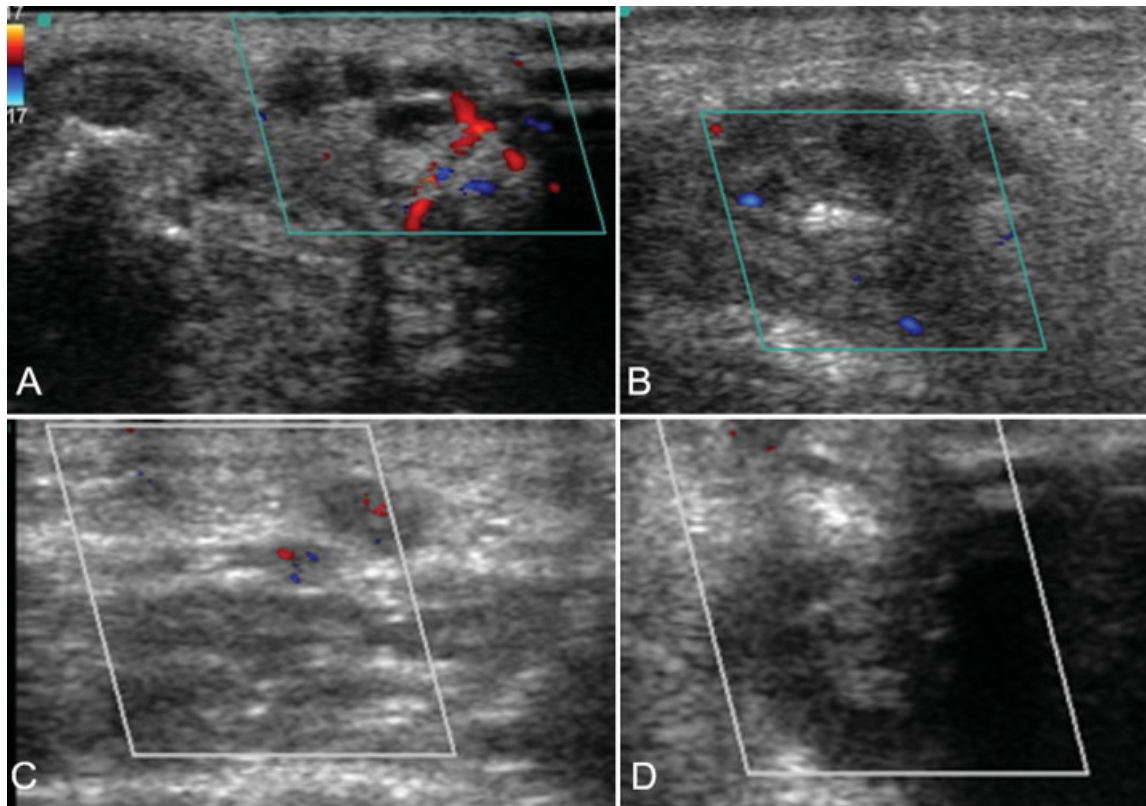


Fig. 6 (A–D) Postembolization 1- (A and B) and 4-month (C and D) follow-up ultrasound of right pinna confirming progressive thrombotic obliteration of AVM nidus with no recurrence.

Conclusion

Auricular AVM is relatively uncommon. Embolization followed by a wide excision and repair is the treatment of choice. Prior ECA ligation to control bleeding AVM presents a therapeutic challenge inciting extensive collateralization and precluding further antegrade catheter access. The purpose of this report was to describe our experience with the antegrade approach of embolizing pinna AVM by catheterizing the collateral vessel after previous ECA ligation and possible added role of percutaneous embolization.

Source of Support

None.

Conflict of Interest

None.

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