Brief Clinical Studies

Title

Auricular arteriovenous malformation with macrotia treated with transcatheter arterial embolization, polidocanol foam sclerotherapy and subsequent otoplasty following resection

Kosuke Ishikawa, MD, PhD [1,2]; Munezumi Fujita, MD, PhD[1,2]; and Satoru Sasaki, MD, PhD[1]

[1]Department of Plastic and Reconstructive Surgery, Center for Vascular Anomalies, Tonan Hospital, Kita 4, Nishi 7, Chuo-ku, Sapporo, Hokkaido 060-0004, Japan

[2]Department of Plastic and Reconstructive Surgery, Faculty of Medicine and Graduate School of Medicine, Hokkaido University, Kita 15, Nishi 7, Kita-ku, Sapporo, Hokkaido 060-8638, Japan

***Corresponding author**: Satoru Sasaki

Department of Plastic and Reconstructive Surgery, Center for Vascular Anomalies, Tonan Hospital, Kita 4, Nishi 7, Chuo-ku, Sapporo, Hokkaido 060-0004, Japan

Tel: +81-11-231-2121, Fax: +81-11-261-8692, E-mail: satoru-s@tonan.gr.jp

Conflicts of Interest and Source of Funding: None.

Abstract

Auricular arteriovenous malformation (AVM) occasionally accompanies macrotia. Here we report a case of AVM with macrotia that was treated with transcatheter arterial embolization, percutaneous sclerotherapy, and subsequent otoplasty following partial resection. A 46-year-old man presented with Schobinger stage III AVM. After transcatheter arterial embolization of the feeding arteries using *n*-butyl-2-cyanoacrylate, 9 sessions of sclerotherapy were performed using 3% polidocanol foam. Partial resection of the AVM nidus and subsequent otoplasty for ear reduction were performed at the age of 50 years. Two years later, the remnant nidus was resected and the protruding ear was surgically corrected. No recurrence was observed, and the enlarged ear was reduced at follow-up 6 months after the final operation.

Keywords

Arteriovenous Malformation; Ear Auricle; Embolization; Sclerotherapy; Vascular Malformation

Introduction

Arteriovenous malformation (AVM) is a lesion related to vascular dysmorphogenesis and consists of an abnormal connection between the arterial and venous system without intervening capillaries, known as a nidus.[1] The most common sites in the facial region are the cheek, ear, lips, nose, and forehead.[2] Auricular AVM usually progresses from Schobinger stage I to stage III with worsening signs and symptoms. A warm cutaneous blush or vascular birthmark is frequently noted on the ear at birth (stage I). With expansion, the auricular skin thickens and its color changes from red to a violaceous hue. The affected ear grows larger with a palpable or audible thrill (stage II), and is eventually accompanied by pain, ulceration, bleeding, and infection (stage III).[3]

Intervention with preoperative embolization and partial or total amputation is recommended if auricular and para-auricular AVM progresses to stage III.[3] Although some authors have described selective embolization with ethanol for auricular AVM,[4-6] improvement following embolization is temporary, lasting at most for a few years.[3] Herein, we report a case of auricular and para-auricular AVM with macrotia that was successfully treated with transcatheter arterial embolization, percutaneous polidocanol foam sclerotherapy, and subsequent otoplasty following resection.

Patient

A 46-year-old man was referred to us with a diagnosis of left macrotia due to AVM. He reported progressive deformity of the left external ear since birth with episodes of auricular bleeding after traumatic abrasion at the ages of 10 and 25 years. On examination, the left external ear was larger than the contralateral ear (length by width, 88 by 48 mm vs 71 by 35 mm), with erythema, tenderness, warmth, pulsation, bruit, and buzzing (Fig. 1A, B). A computed tomography (CT) scan with contrast enhancement revealed a strongly enhancing soft tissue mass around the left ear that extended to the ipsilateral parotid gland and cervical subcutaneous region (Fig. 1C). Selective angiography showed dilation and tortuosity of the feeding arteries, mainly from the posterior auricular artery, and draining veins to the external jugular vein. He was diagnosed with Schobinger stage III AVM and treatment was planned to preserve the ear.

First, transcatheter arterial embolization of 2 main feeding arteries was performed using *n*-butyl-2-cyanoacrylate, resulting in successful embolization of the arteriovenous fistulae in the retroauricular region. Three days later and again 4 months later, 2 sessions of percutaneous sclerotherapy were performed under general anesthesia using 20 mL of polidocanol foam mixed with indocyanine green under duplex ultrasound and near-infrared

fluorescence imaging guidance[7] in the auricular region (Supplemental Digital Content). Six further sessions of percutaneous sclerotherapy were performed using 10 mL of polidocanol foam under duplex ultrasound in the outpatient setting at 3- to 6-month intervals. A ninth session of percutaneous sclerotherapy was performed using 50 mL of polidocanol foam under general anesthesia. A year later, we performed partial resection of the AVM nidus and subsequent otoplasty for ear reduction at the age of 50 years.

Partial resection of the AVM nidus was performed in the base of the auricular lobule (Fig. 1D, E). A falciform anterior skin excision was made along the sulcus of the helix from the top to the middle of the scapha, and the underlying cartilage was transected to reduce width while leaving the posterior skin intact. A full-thickness resection of 1 cm in height was made in the helix and scapha at the level of the crus of the helix, continuing to a retroauricular incision. Next, a stair-step anterior skin incision of 1 cm in height was made in the antihelix with the underlying cartilage transected to reduce height. A crescent-shaped anterior skin incision in the posterior wall of the concha was made with the underlying cartilage transected to reduce projection. Finally, a crescent-shaped anterior skin incision was made along the sulcus of the helix at the lobule to reduce height. Sutures were placed along anatomical structures except 2 horizontal lines across the helix and antihelix (Fig. 1F).

Two years later, we resected the remnant nidus in the retroauricular region and surgically corrected the protruding ear by resection of the conchal cartilage and its plication. There was no recurrence, and the enlarged ear was reduced at follow-up 6 months after the final operation (Fig. 2A, B). A postoperative CT scan with contrast enhancement demonstrated reduction of the tortuous vessels in the external ear with persistence of a dilated external jugular vein (Fig. 2C).

Discussion

Intervention is unnecessary for patients with stage I to II auricular AVM but should be considered upon progression to stage III.[3] Wu et al reported that only preoperative embolization and partial or total amputation could control auricular and para-auricular AVM.[3] However, some studies have reported cure with selective ethanol embolization alone in 18% to 75% of patients with auricular AVM.[4-6] Those studies also reported reversible skin necrosis in 11% to 30% of patients as a complication.[4-6] However, disfigurement caused by macrotia was not improved by ethanol embolization alone even in a patient who was cured.[4] Although the sclerosing effect of polidocanol is more moderate than that of ethanol, it can be used to treat AVM.[8]

Ear reduction seems to be the least popular of operations involving the external ear. Full-thickness wedge resection is the simplest method, but a direct suture to close the defect causes cupping deformity of the ear because the cartilage is not stretchable. Argamaso reported a technique for ear reduction in which a falciform excision is made along the sulcus of the helix, resulting in a less obtrusive scar line that stays within the helical groove; the redundancy of the helix was corrected by resecting a portion of it near the crus or at the lobule.[9] We performed otoplasty for ear reduction in combination with stair-step resection in the middle of the ear and falciform excision along the sulcus of the helix in the upper half of the ear and the lobule because blood supply to the helix might have been reduced by multiple sclerotherapy sessions. Cruciform scars across the helix and antihelix were minimized by staggered transection of underlying cartilage with respect to the skin above. Given complete resection of the auricular AVM is difficult without amputation, long-term follow-up is needed with maintenance sclerotherapy.

References

1. Liu AS, Mulliken JB, Zurakowski D, et al. Extracranial arteriovenous malformations: natural progression and recurrence after treatment. *Plast Reconstr Surg* 2010;125:1185-1194
2. Kohout MP, Hansen M, Pribaz JJ, et al. Arteriovenous malformations of the head and neck: natural history and management. *Plast Reconstr Surg* 1998;102:643-654
3. Wu JK, Bisdorff A, Gelbert F, et al. Auricular arteriovenous malformation: evaluation, management, and outcome. *Plast Reconstr Surg* 2005;115:985-995
4. Jin Y, Lin X, Chen H, et al. Auricular arteriovenous malformations: potential success of superselective ethanol embolotherapy. *J Vasc Interv Radiol* 2009;20:736-743
5. Zheng LZ, Fan XD, Zheng JW, et al. Ethanol embolization of auricular arteriovenous malformations: preliminary results of 17 cases. *AJNR Am J Neuroradiol* 2009;30:1679-1684
6. Wang D, Su L, Han Y, et al. Ethanol embolotherapy of high-flow auricular arteriovenous malformations with electrolytically detachable coil-assisted dominant outflow vein occlusion. *Eur J Vasc Endovasc Surg* 2014;48:576-584
7. Ishikawa K, Sasaki S, Furukawa H, et al. Preliminary experience with intraoperative near-infrared fluorescence imaging in percutaneous sclerotherapy of soft-tissue venous malformations. *Dermatol Surg* 2013;39:907-912
8. Ergun O, Atli E, Gulek B, et al. Intraarterial polidocanol injection for the treatment of peripheral arteriovenous malformations. *Surg Today* 2014;44:1232-1241
9. Argamaso RV. Ear reduction with or without setback otoplasty. *Plast Reconstr Surg* 1989;83:967-975

Figures

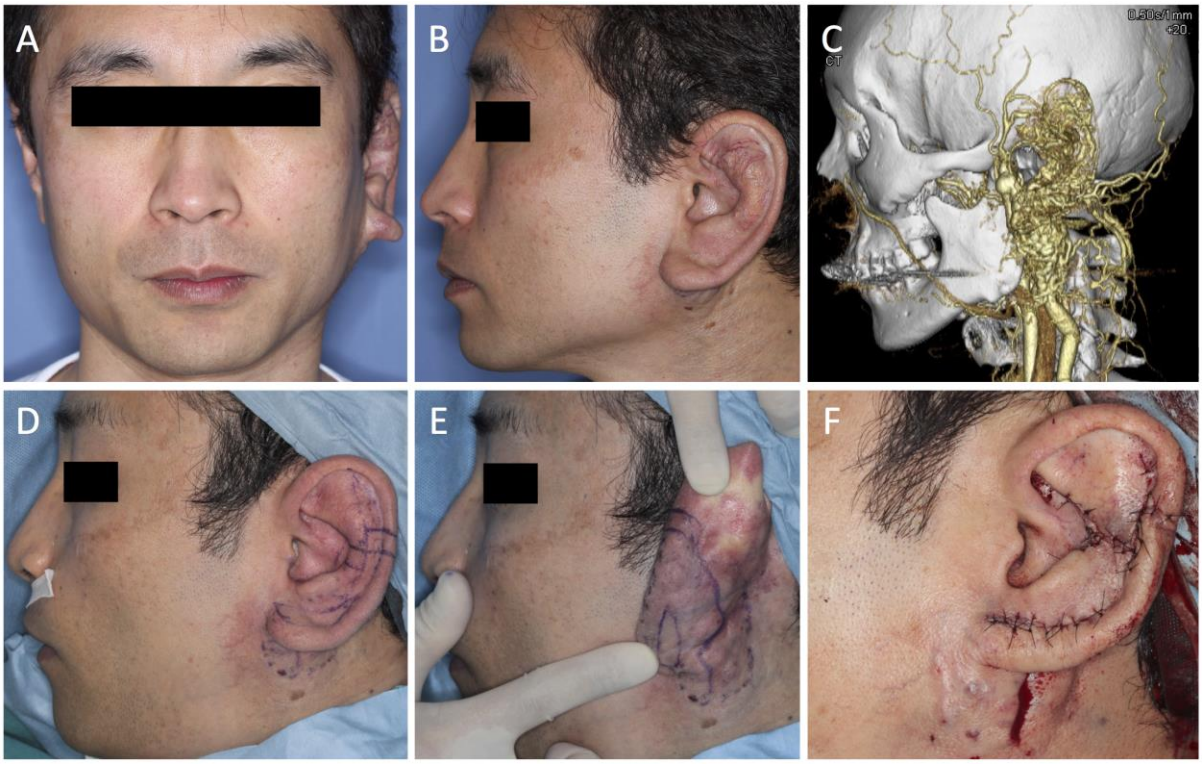

Figure 1. (A, B) Photographs taken at the first clinic visit show an enlarged left ear. **(C)** Enhanced computed tomography scans before treatment show a lesion around the left ear extending to the ipsilateral parotid gland and cervical subcutaneous region. **(D, E)** Design of the incision for partial resection of the arteriovenous malformation and otoplasty. **(F)** Intraoperative view after otoplasty.

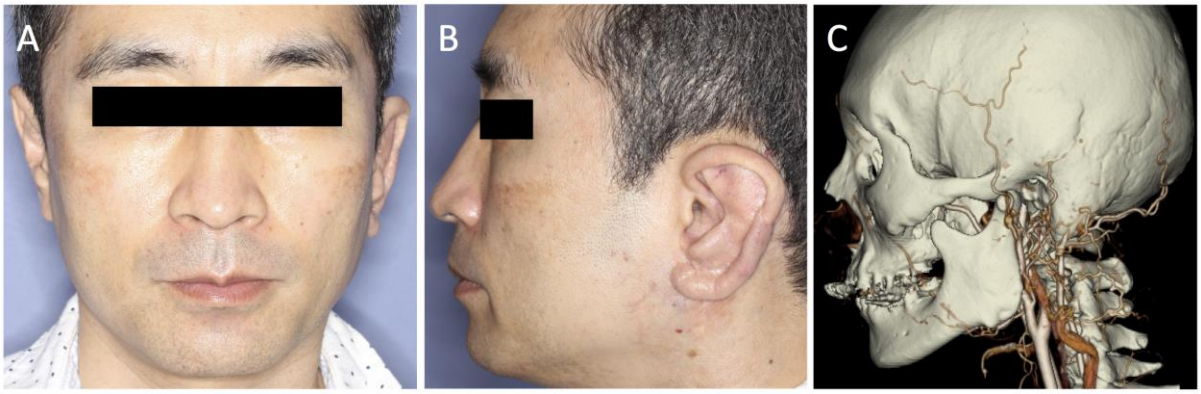

Figure 2. Findings at 6 months after the final operation. **(A**, **B)** Photographs show no recurrence with reduction of the enlarged left ear. **(C)** Enhanced computed tomography scans show reduction of most of the tortuous vessels in the external left ear with a dilated external jugular vein.

Supplemental Digital Content. Percutaneous sclerotherapy using polidocanol foam mixed with indocyanine green under duplex ultrasound and near-infrared fluorescence imaging guidance in the auricular region.

http://links.lww.com/SCS/C324