

Midterm and long-term results of ethanol embolization of auricular arteriovenous malformations as first-line therapy

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ABSTRACT

Objective: The objective of this study was to assess the midterm and long-term results of patients with auricular arteriovenous malformations (AVMs) treated by ethanol embolization.

Methods: Medical records of 35 patients (20 male, 15 female; age range, 10-59 years) with auricular AVMs between 2006 and 2016 were reviewed. The short-term results of 4 of the 35 patients were reported in 2009. They were included in this study with updated follow-up data. The data from the 31 new included patients have not been reported previously. Ethanol embolization was performed with a direct puncture approach. The nidus was eradicated by bolus injection of ethanol with manual compression whenever possible. Treatment outcomes were classified into four categories by assessing the resolution of symptoms as well as the degree of nidus devascularization between the baseline and follow-up angiography studies.

Results: A total of 86 embolization sessions were performed in 35 patients (mean, 2.5 ± 1.3 sessions). The dosage of ethanol used per single session was 12.0 ± 4.5 mL (range, 6.0-24 mL). All patients received post-treatment clinical follow-up (mean, 40.7 ± 25.8 months), and 28 patients received post-treatment imaging follow-up (mean, 34.3 ± 25.5 months). Ethanol embolotherapy was effective in all patients. Control was achieved in 16 patients (45.7%), and improvement was achieved in 18 patients (51.4%). One patient experienced recurrence. A total of 13 minor complications and 2 major complications occurred in 12 patients during the 86 treatment sessions (12/35 [34.2%]; 15/86 [17.4%]). All the complications resolved spontaneously.

Conclusions: The midterm and long-term results of this study demonstrate that ethanol embolization alone is an effective option for auricular AVMs as first-line therapy with a mild risk of minor and major complications. (*J Vasc Surg: Venous and Lym Dis* 2018;■:1-10.)

Keywords: Arteriovenous malformations; Ethanol; Embolization

Arteriovenous malformation (AVM) is the rarest congenital vascular malformation subtype, with a high-flow shunt between the feeding artery and the outflow vein without a normal capillary network.¹ The ear is the second most common site for extracranial AVMs in the head and neck region.² Located in such a precise aesthetic and functional region as the head and neck, auricular AVMs are distinctive, compared with other peripheral AVMs, because lesion expansion often results in deformities that are usually not life-threatening but can cause substantial aesthetic and social disturbances.³⁻⁵

Historically, complete excision of an AVM lesion is ideal and is considered the only treatment of choice.^{2,6} However, one cannot attempt radical excision without considering postoperative functional and aesthetic damage, especially when the lesion is located in the auricular region. Resection and reconstruction often result in a more noticeable deformity than the malformation itself.⁷ Furthermore, as an "extratruncular" lesion resulting from defective development in its early stage of embryogenesis, the lesion carries mesenchymal cell characteristics to grow. Even after radical resection and reconstruction, AVMs may recur.^{2,6,8} A large series of 272 patients with head and neck AVMs showed an 81% recurrence rate after surgical resection and a 98% recurrence rate after embolization.⁶ Goldenberg et al⁸ published a study on the surgical resection of extracranial AVMs after presurgical embolization. In that series, the overall regrowth rate in the 31 patients was 32.2% (10 of 31 patients), but 18.2% of the patients experienced recurrence even after total resection, with a mean follow-up of 3.5 years.

Ethanol embolization is theoretically able to provide curative outcome when it is done properly and can achieve obvious shrinkage of the AVM lesions.^{4,9} Although multiple studies have been published on the ethanol embolization of AVMs, literature regarding

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the long-term results of the ethanol embolization of auricular AVMs is lacking. These anomalies are often included in large series of head and neck AVMs. To our knowledge, only three small case series^{4,10,11} have reported ethanol embolization for auricular AVMs and have demonstrated good short-term clinical success. However, the treatment efficacy of ethanol embolization of auricular AVMs in a larger cohort has not yet been reported, and the medium-term and long-term results are unknown.

This study was designed to update the long-term results of patients with auricular AVMs treated by ethanol embolization reported in 2009⁴ and to report the outcomes of patients treated with the same technique from 2009 to 2016.

METHODS

The study was approved by the Institutional Review Board, and all subjects gave informed consent. The medical records of 35 patients (20 male and 15 female) with auricular AVMs between 2006 and 2016 were reviewed and assessed. The ages of the patients ranged from 10 to 59 years (mean, 29.4 years).

The short-term (4-27 months) results of 4 of the 35 patients were reported in a study of auricular AVMs of 8 patients in 2009.⁴ Of these eight patients, four patients have since been lost to follow-up; the remaining four patients were included in this study with updated follow-up data. The data of the 31 new patients (1 treated before 2009 and 30 treated after 2009) have not been reported previously.

Seventeen patients (48.6%) had undergone previous treatments (no application of ethanol): surgery (n = 7), sclerotherapy (n = 4), embolization (n = 3), laser (n = 2), surgery with embolization (n = 1), and cryotherapy (n = 1) or ligation of the proximal supplying arteries (n = 1). No response (n = 5) and deterioration of symptoms (n = 12) were reported by those patients.

The AVM was diagnosed on the basis of clinical findings and imaging studies including magnetic resonance imaging (MRI; n = 25), computed tomography angiography (CTA; n = 27), and color flow Doppler ultrasound imaging (n = 13). All initial diagnoses were confirmed by digital subtraction angiography (DSA) as the "gold standard" for diagnosis. Two surgeons evaluated the symptoms and agreed on the clinical stage according to the Schobinger grading system.²

Endovascular procedures. All procedures were performed under general anesthesia. Detailed DSA was performed to depict the angioarchitecture of the nidus according to the Cho-Do¹² classification system (Fig 1).

In all patients, the nidus of the lesion was accessed with a direct puncture approach. Before puncture, the location of the nidus was estimated by analyzing prior MRI or CTA findings and confirmed by detailed DSA.

ARTICLE HIGHLIGHTS

- **Type of Research:** Retrospective cohort study
- **Take Home Message:** In 35 patients with auricular arteriovenous malformations treated by 86 ethanol embolization procedures, excellent to good results were seen in 34 patients, with one recurrence. Fifteen complications resolved spontaneously.
- **Recommendation:** Ethanol embolization is an effective treatment for auricular arteriovenous malformations.

The access to the nidus was then comprehensively determined by understanding the anatomic position of the nidus relative to adjacent structures.

Direct puncture was performed with a 21-gauge needle (Surflo Winged Infusion Set, 21-gauge, 0.75-inch; Terumo, Leuven, Belgium), which was advanced forward and backward to detect the nidus according to imaging analysis. When fast blood reflux was noted, which suggests that the needle tip is intraluminal within a corresponding nidus vessel, angiography was performed to confirm the location of the needle tip. Keeping the needle tip immobile during manipulation is crucial because any movement will dislodge the needle tip from the tiny vessel lumen. An optimal puncture site was confirmed only when opacification of the nidus and the draining veins, but not the feeding arteries, was evident under angiography.

Test injection with contrast material was performed as a reference for the injection rate and the volume of the ethanol used during the subsequent embolization procedure. The nidus was eradicated by bolus injection of ethanol with manual compression of the draining veins whenever possible with a total amount not exceeding 0.4 mL/kg.⁴ When the maximum tolerated dosage of 0.14 mL/kg during 10 minutes was met,¹³ a 5-minute waiting period was necessary. Repeated arteriography was performed after each injection to delineate the real-time changes in the hemodynamics and angioarchitecture of the lesion to determine whether to continue the injection at the present puncture site. If the contrast agent was observed leaking out of the nidus into adjacent tissues or disappearance of the draining veins and persistent arterial reflux occurred, which indicate a risk of nontarget embolization, the current puncture site was considered unsafe and abandoned. In this circumstance, we punctured another site of the lesion and sought a new optimal injection site by repeating the aforementioned procedure. Usually, absolute ethanol was used; however, in cases of infiltrative lesions (infiltrative patterns of the nidus constituted by micro-fistulas), which mainly involve the dermis and carry a high risk of skin necrosis, diluted ethanol (50%-75%

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