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Original Article

Surgical Treatment with or without Embolotherapy for Arteriovenous Malformations

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Objective: The management of arteriovenous malformations (AVMs) remains challenging due to the high rate of recurrence of these lesions. Surgical resection is the only potential cure; however, it is often difficult to perform and carries a risk of massive hemorrhage. The purpose of this study was to review our experience with AVMs treated by surgical resection.

Materials and Methods: We retrospectively reviewed the medical records of nine patients with AVM, treated with surgical resection. We treated these patients with excision surgery with or without embolotherapy.

Results: Eight were treated with surgical resection with embolotherapy and one was treated with a simple surgical resection. Five patients with AVMs were cured. However, two cases of AVM recurred after total excision, and AVMs remained in two cases of partial excisional surgery in which the lesions involved the joints.

Conclusions: Total excision of AVMs leads to a cure; however, total excision is not adequate in cases of AVMs involving the joints. Multidisciplinary treatment may offer good results in reducing the morbidity. To minimize complications related to surgery, aggressive control of blood flow to the lesion, preoperatively, with appropriate embolotherapy is essential, and a complete resection with a chance of cure will be increased.

Keywords: arteriovenous malformations, surgical resection, embolotherapy

Introduction

Arteriovenous malformations (AVMs) are rare congenital lesions characterized by large numbers of arterial feeding branches.[1] Complete eradication of the nidus of an AVM is the only potential cure.[2] However, surgical resection is often difficult and carries the potential for causing exsanguinating hemorrhage and damage to the surrounding structures. Recurrence of AVMs is common following incomplete resection. Ligation of the large feeding vessels should be avoided because it makes subsequent embolization more difficult and is never curative, but rather, it stimulates its mesenchymal cell characteristics, and the lesion gets worse by neovascular recruitment.[3] Accordingly, transcatheter embolization now plays a significant role in the treatment of patients with AVMs.[3] Performing preoperative embolization prior to surgical intervention may be helpful in order to reduce the risk of massive bleeding. When treatment is indicated for a lesion, it should be directed at eradicating the nidus of the lesion with embolization and/or surgery.

The aim of this study was to review our clinical results of surgical treatment of AVMs, with or without embolotherapy, and evaluate patient outcomes.

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Table 1 Treatment indications for arteriovenous malformations

Absolute indications
Hemorrhage (3/9)
Gangrene or ulcer (0/9)
Cardiac failure (0/9)
Relative indications
Various symptoms and signs affecting the quality of life; pain and/or functional impairment (7/9)
Lesions with potentially high risk of complications and/or limb-threatening location (2/9)
Limb length discrepancy (0/9)
Cosmetically severe deformity (1/9)

Demographic data of 9 patients described in this study are included

Materials and Methods

All patients provided their informed consent. Between January 1992 and December 2011, nine patients underwent surgical excision with or without embolotherapy to treat AVMs at Tokyo Medical and Dental University Hospital. Patients who underwent only embolotherapy were excluded from the study. The data were obtained retrospectively via a thorough and extensive review of all medical records. Among the nine patients, there were two males and seven females with a mean age of 42 years (age range: 25–67 years).

Clinical diagnosis of AVM was made based on the findings of a physical examination. Non-invasive tests, such as duplex ultrasound imaging, computed tomography (CT) and magnetic resonance imaging (MRI), were used to make definitive diagnoses and assess the extension of the AVMs, especially those extending into the muscles, bones or joint space. Next, selective catheter contrast angiography was performed to confirm the location of the feeding arteries and arteriovenous fistulae and create a therapeutic plan.[4)]

Treatment indications are described in **Table 1**. The majority of patients had various symptoms affecting their quality of life, such as pain (77%). Absolute indications with hemorrhage were recognized in three patients.

To minimize adverse effects related to surgery, such as massive bleeding, the use of embolotherapy was considered preoperatively. The selection of embolotherapy as adjunct therapy administered before surgical therapy depended on the location and extent of the lesions. Surgical procedures were preferred for all accessible AVMs associated with an acceptable risk of attempting a complete cure. As embolic agents, ethanol

Table 2 Symptoms and signs of surgically treated arteriovenous malformations

Symptoms and signs	*n*(%)
Thrill and pulsation	8 (88)
Pain	7 (77)
Bleeding	3 (33)
Sensation of heat	2 (22)
Cosmetic problems	1 (11)

N - butyl cyanoacrylate (NBCA) (Histoacryl®, Aesculap AG, Tuttlingen, Germany), various types of coils and Gelfoam pledgets were used in various combinations, either simultaneously or in stages, depending upon the location, severity and extent of the AVM. NBCA glue was used primarily for surgically excisable lesions as preoperative embolotherapy to reduce morbidity during subsequent surgical therapy.[2)]

Treatment responses, as well as interim and final results, were assessed periodically. Clinical assessments were based on improvements in clinical symptoms. Laboratory assessments were based on various modalities, including ultrasonography, CT and MRI. During the follow-up period, if a recurrence of an AVM were observed, we would perform further therapy, surgical resection and/or embolotherapy.

Results

We treated nine patients with AVMs. Eight patients with AVMs were treated with surgical excision in addition to embolotherapy. One patient was treated solely with surgical excision. Nine patients who underwent surgical treatment showed various signs and symptoms such as thrills, pain and bleeding, which are summarized in **Table 2**.

The patients with surgically treated AVMs included two patients with shoulder AVMs, two patients with thigh AVMs, two patients with foot AVMs and one patient each with AVMs of the back, buttocks and fingers. Of the nine cases, four (two cases of shoulder AVMs, one case of a thigh AVM and one case of a foot AVM) involved the joints.

Locations and treatments of the AVMs Table 3

Back: The patient was firstly treated with embolotherapy using Gelfoam and ethanol. After one month, partial excision of the nidus was performed. Two months after the first excisional surgery, second

Table 3 Treatment by the location

Location	Treatment (In order of therapy)	Outcome
Back	E - S - E - S	Cure
Shoulder	E - S	Recurrence
	S - E - L	Remaining
Buttock	E - S - E	Cure
Finger	L - L - L - S - S	Amputation
Thigh	S	Remaining
	E - E - S	Cure
Foot	L - S - S - E - E - L	Recurrence
	S - S - E - E - S	Cure

E: embolotherapy; S: surgical resection; L: ligation

embolotherapy was performed with coil embolization. One week later, total excision of the nidus was performed. No recurrence was observed during 75 months of follow-up after surgery.

Shoulder: One patient underwent embolotherapy with Gelfoam and coil embolization. One week later, total excisional surgery was performed. Sixty-eight months after surgery, a recurrence of the lesion was observed; however, the patient's pain was mild, and the patient did not wish further treatment; then no further treatment was performed. The other patient underwent primary, partial excision surgery, and the patient complaint was relieved. One year after the first surgery, remnant lesion grew up, then embolotherapy with Gelfoam and coil embolization was performed. One month later after the embolotherapy, the mass was not reduced, so ligation of the branches of the avillary artery was performed. While the AVM could not be completely resected, the patient's pain was relieved. All of these cases received follow-up with no complaints.

Buttock: The patient was treated with preoperative embolotherapy using Gelfoam, and one month later, total excisional surgery was performed. Seventy-six months after surgery, a recurrent AVM was observed in the same lesion, and adjunct embolotherapy with Gelfoam and coil embolization was performed, thus resulting in no obvious recurrence for 79 months.

Finger: The patient was treated with three ligation surgeries; however, the AVMs progressed. Therefore, 3 years after the first ligation surgery, amputation surgeries were required twice, and the thumb and second finger of the left hand were amputated. No obvious recurrence was observed during 24 months of follow-up after the amputation surgery.

Thigh and/or calf: One patient was treated primarily with excisional surgery. The AVM lesion involved the knee joint; therefore, we performed partial excision. After surgery, symptoms of bleeding were controlled; however, venostasis occurred in the calf. The other patient was admitted to our hospital for massive bleeding, and emergency embolotherapy using NBCA was performed, which led to hemostasis. Two weeks after the first embolotherapy, adjunct embolotherapy with NBCA was performed. Two weeks after the second embolotherapy, total excision could be performed because the lesion was continuous with the thigh and did not extend to the vessels or joint. No clinical signs of recurrence were recognized on MRI during 14 months of follow-up after surgery.

Foot: The left dorsal pedis artery was ligated, and the AVM was excised completely during twice-staged surgery in a patient with an AVM located in the left side of the ankle. Six months after surgery, recurrence was recognized in the left ankle. Therefore, first embolotherapy with Gelfoam and coil embolization were performed, then one month later, adjunct second embolotherapy using Gelfoam and coil embolization performed. One week after the second embolotherapy, ligation of the feeding artery were performed. Even though the lesion remained and required partial treatment, the patient's pain was relieved. The other patient was treated with two excisional surgeries. Fourteen months after the second surgery, a recurrent lesion was revealed in the right heel. Therefore, two preoperative embolotherapies with Gelfoam and coil embolization were performed, and 6 months after the second embolotherapy, the recurrent lesion was excised completely without any signs of recurrence 171 months after the previous treatment.

Case presentation

We demonstrate one case of a patient who underwent surgical treatment, in addition to preoperative embolotherapy, in **Fig. 1**. A 37-year-old female with an AVM in her left thigh **Fig. 1a** was admitted and underwent two preoperative embolotherapies **Fig. 1b, 1c and 1d**. Following surgery, the AVM was resected completely, and the defect was covered with a full-thickness skin graft **Fig. 1e**. Postoperatively, the patient achieved good results regarding wound healing, and no recurrent lesions were observed 11 months after surgery.

Discussion

AVMs are usually present at birth; however, not all of them are easily detectable clinically.[5] Signs and

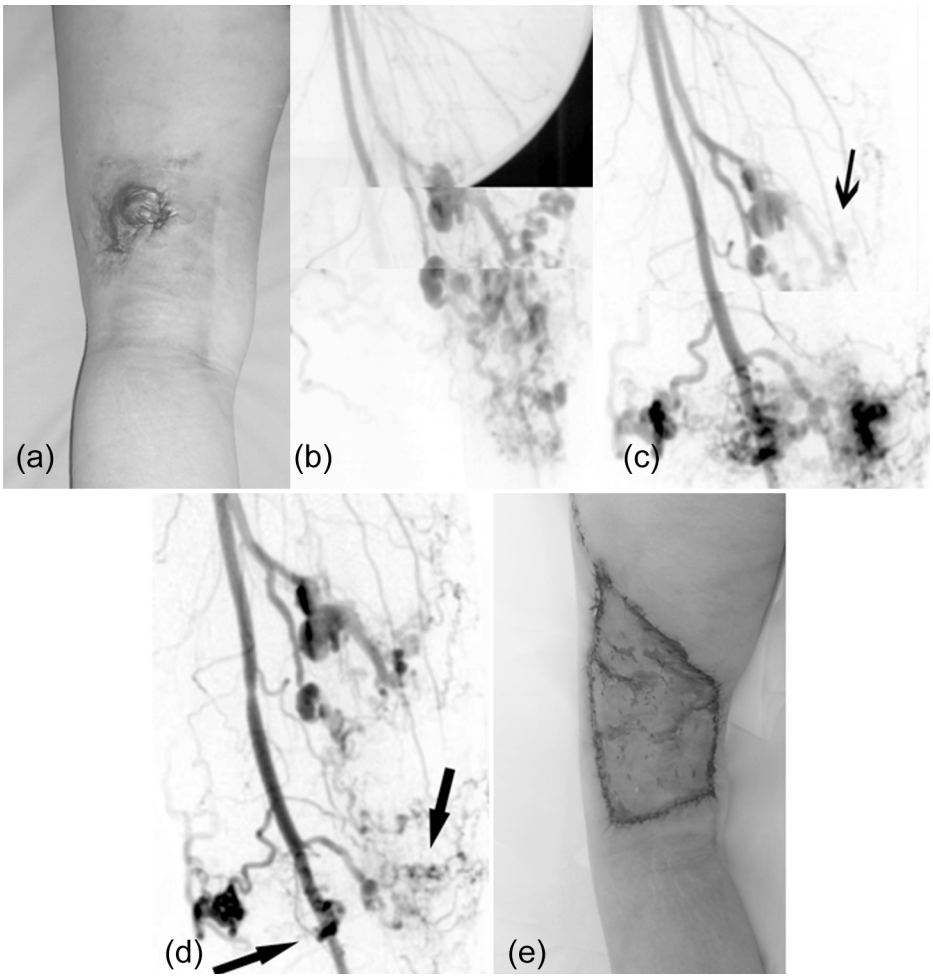

Fig. 1 **(a)** The preoperative gross findings showed an AVM in the left thigh. **(b)** The AVM in the branches of the deep femoral artery was revealed pre-embolotherapy. **(c)** After the first embolotherapy, the nidus of the AVM was reduced in size (black arrow). **(d)** After the second embolotherapy, angiogram showed a reduction in the size of the nidus (black arrows). **(e)** After surgery, the AVM was cured with a skin graft.

symptoms become manifest later in life. Some lesions become apparent after trauma or during periods of hormonal stimulation such as menarche or pregnancy.[6)]

The diagnosis of congenital AVM can usually be made according to the patient's history and a physical examination. Confirmation is generally obtained with CT scanning, MRI or angiography. Performing MRI is not only valuable for making the diagnosis, but it may also demonstrate treatment responses during the follow-up period. If the nidus of the AVM is subcutaneous or has partially invaded the muscle, the AVM can be completely removed. Therefore, CT or MRI demonstrate whether an AVM is resectable. Imaging is also useful in cases with multiple lesions and can identify complications, including hemorrhage.[7)] Angiography is the gold standard tool for identifying the feeding arteries and flow status of arteriovenous fistulae.

Treating AVMs is generally indicated in cases of enlarging lesions, and it should be performed before significant overgrowth, or if severe disfigurement of the patient develops. Other symptoms requiring treatment include bleeding of the lesion, critical ischemia, disabling pain and, rarely, congestive heart failure.

AVMs present an extremely difficult therapeutic challenge for both surgical and nonsurgical treatment modalities. Performing complete resection is extremely challenging and can be problematic or even catastrophic. Surgical treatment alone can cause massive bleeding during surgery.[8)] However, whether AVMs

are resectable depends on their location. If an AVM involves the joints, ligaments or vessels, we should consider whether total resection and reconstructive surgery would perform or not. In our series, two cases involved the shoulder or knee joints. In these cases, we could not avoid performing only partial excision due to the severe invasiveness of the AVMs in the joint space. In the knee joints involvement case, the nidus was fed from the superficial femoral artery, which caused hemorrhaging into the knee joints, so we performed excision surgery to control the hemorrhage. Intraoperative findings showed that the nidus was close to the superficial femoral vein, so we performed partial resection of the superficial femoral vein and reconstructive surgery. Two patients had been treated with total excision in the shoulder or foot; however, the AVMs reappeared. Even though it was difficult to treat and manage the AVMs, including those in the joints, partial excision contributed to the relief of the patients' symptoms,[9] thus resulting in careful surveillance and prompt intervention and/or surgery.

Preoperative embolization is useful and effective for reducing the incidence of intraoperative hemorrhage, thereby improving surgical outcomes and decreasing the recurrence rate in the long term.[7] In our series, we performed preoperative embolotherapy prior to surgery in one out of two thigh cases. One patient was treated with primary excision, and the amount of intraoperative blood loss in that case measured 3,632 ml. The other patient was treated with total surgical resection and preoperative embolotherapy, and the amount of intraoperative blood loss in that case measured 712 ml. Therefore, preoperative embolotherapy can be useful for reducing the amount of intraoperative blood loss. Numerous embolic materials have been developed, ranging from Gelfoam pledgets to complex systems employing microcatheters and detachable coils. NBCA cannot act as a permanent agent to control lesions effectively in cases without evidence of permanent damage to the endothelium.[10] Ethylene-vinyl alcohol copolymer (Onyx®, Micro Therapeutics, CA, USA) might be used in such cases instead of NBCA. Onyx®, unlike other liquid embolic agents, does not adhere to the endothelial wall or catheter tip.[11] Onyx® is less adhesive than NBCA; therefore, we prefer NBCA to Onyx®. Platinum coils are permanent embolic materials used to occlude proximal sites and control blood flow.

The current management of AVMs is based on a new concept of a multidisciplinary approach[12,13] that can minimize morbidity and reduce recurrence. The limited role of embolotherapy as an adjunctive therapy for surgical resection has been further expanded. This approach has even been helpful in treating high-risk lesions with a high flow status. The importance of making a careful assessment of the treatment strategy based on the ratio of the benefits to the potential risks cannot be overemphasized.[12]

Conclusion

Treating AVMs is a challenging issue for vascular surgeons. Multidisciplinary treatment may offer a better result. In order to excise an AVM completely, reconstructive surgery may be needed. To minimize complications related to surgery, aggressive control of blood flow is essential, and complete resection may be required.

Disclosure Statement

Dr. Igari and the other co-authors have no conflicts of interest to declare.

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