Treatment for congenital arteriovenous malformation (AVM) of peripheral vessel is a challenge. In unresectable AVM, embolization is an alternative treatment modality. We report two cases of large AVMs at lower extremities. Transcatheter arterial embolization (TAE) was done after attempted surgery which was complicated by persistent wound oozing and poor wound healing. Polyvinyl alcohol particles and gelfoam pledgets were used as embolizing materials. The outcome is satisfactory without complication.

Key words: Arteriovenous malformation; Extremities, transcatheter arterial embolization

Large peripheral congenital arteriovenous malformation (AVM) is very difficult to manage. Surgical cure is uncommon, and deep lesion requires amputation or is unresectable. Transcatheter arterial embolization (TAE) targeted at obliteration of the core nidus may provide a safe and efficacious therapeutic method in selected group of patients when the lesions are not resectable.

With recent refinement in catheter system and digital vascular imaging techniques, issue of inadvertent non-target tissue embolization decreases. Multi-stages TAE plays an important role in AVM management.

Herein we report two cases of huge AVMs at proximal portion of lower extremities, which were successfully treated by TAE.

CASE REPORTS

Case I

An 18-year-old male patient found a small painful nodule on his right buttock. This lesion progressively enlarged for 4-5 months. Hence, he came to our hospital for help. The lesion was about 6x6 cm in size with skin defect and ulceration. Engorged vessels and difficulty in hemostasis were appreciated at excisional biopsy. Poor wound healing was observed also. Pelvic CT reveals an ill-defined, soft tissue lesion about 4x5x10 cm3 in size in right buttock and proximal thigh. Post contrast CT shows mild enhancement of the lesion and tortuous tubular structures in adjacent subcutaneous fat. Hypertrophy of the right gluteus maximus muscle was also noted. Angiography of the right common femoral artery shows complex nidus, engorged arteries and veins in the right gluteal region (Fig.1A). AVM was diagnosed according to the imaging findings. Branches of right superficial and deep femoral arteries supply this lesion. Engorged serpentine veins are also identified.
Due to persistent wound oozing and poor wound healing after attempted surgical resection, TAE was carried out via contralateral femoral artery approach with a 4F RC1 catheter (Cordis Europa N.V., Roden) selectively engaged in the bigger feeding arteries from the branches of deep femoral artery. Polyvinyl alcohol (PVA) particles of 300 to 600 µm (Ultra-Drivalon, Nycomed Amersham medical system, Paris) were introduced until slowdown of the forward flow, then Gelfoam pledgets (Pharmacia & Upjohn Company, Kalamazoo, MI) were subsequently injected to completely devascularize the AVM (Fig.1B). The 2nd and 3rd TAE (Figs.1C, 1D) were performed 3 months and 6 months later, respectively, via superselective catheterization of the same feeder branches leading to the residual AVM. Dramatic shrinkage of the mass-like lesion and improvement in wound healing were appreciated gradually. Wound healing was complete and no symptom remained at follow-up for more than one year.

Case II

An 18-year-old male patient found a nodule in the left inguinal area, and this lesion increased in size progressively within a period of 8 months. Due to spontaneous bleeding, he came to our ER for help. In physical examination, this lesion was 12 × 17 cm in size with cutaneous ulceration. Pelvic CT reveals an ill-defined bulging mass about 10 × 16 × 15 cm in left lower pelvis and subcutaneous tissue of medial side of

**Figure 1.** Case 1. a. Right common femoral arteriography showed an AVM (arrows) over buttock with blood supplies from dilated and tortuous branches of deep femoral artery. The tip of the catheter is marked (arrowhead). b. Status post-1st TAE. A major AVM feeder from right deep femoral artery was obliterated by PVA and Gelfoam pledgets (arrow). c. Six months later, the size of AVM was diminished and the feeding artery was narrowed. d. Status post 3rd TAE. Right deep femoral arteriography showed complete devascularization of arterial feeders (arrow).
the left thigh. Heterogeneous enhancement is identified in the bulging mass and adjacent area on post-contrast CT scan. The bony structure was intact. The left gracilis muscle was hypertrophic. On MRI, the mass is slightly hyperintense on T1WI and very hyperintense on T2WI. Multiple signal-void serpentine tubular structures in both T1WI and T2WI with strong enhancement are also noted. AVM was considered based on imaging findings. Angiography of the left common iliac artery shows a large AVM supplied mainly by engorged left internal pudendal and obturator arteries and their branches. The right obturator artery and perforator muscular branches of the left superficial femoral artery also contributed to this lesion. The lesion was partially removed, but poor wound healing and repeated infection were noted. Angiography was, therefore, performed again with a 4Fr. Cobra catheter (Torcon NB, Cook Incorporated, Bloomington, IN) (Fig.2A). TAE was carried out subsequently (Fig. 2B) via the left obturator artery and internal pudendal arteries. PVA particles (Contour, Boston Scientific/Target vascular system, Fremont, CA.) of 150–250 and 355–500 µm were initially introduced to obliterate the core nidus. Proximal devascularization of the arterial feeders to the AVM was obtained by Gelfoam pledgets administration. Because this AVM was quite large, multi-stages embolization was planned to obviate the possible complications. The mass shrank dramatically after embolization and wound healing was improved. No sexual dysfunction was noted objectively. However, this patient thought that his problem had been resolved, and he did not want to receive second TAE. He didn’t have follow-up after first TAE.

DISCUSSION

Congenital vascular lesion can be divided into two major groups: hemangiomas and vascular malformations. They represent different responses to treatment and the prognoses are also different. The diagnosis is based on clinical history and imaging findings. Hemangiomas are benign vascular neoplasms in infancy caused by endothelial hyperplasia. Most of them regress without treatment by 12 years of age. On angiography, hemangiomas represent irregularly stained vascular channels with lobular pattern. Large feeding arteries will not be visualized. This is the key distinction between AVMs and hemangiomas [1]. In contrast, vascular malformations do not demonstrate endothelial cell proliferation. On angiography, hemangiomas represent irregularly stained vascular channels with lobular pattern. Large feeding arteries will not be visualized. This is the key distinction between AVMs and hemangiomas [1]. In contrast, vascular malformations do not demonstrate endothelial cell proliferation. On angiography, they represent complex vascular nidus, dilated and tortuous feeding arteries and draining veins [2].

Management of a congenital AVM is difficult. Cosmetic disfiguring may result from operation, and if the lesion involve muscle groups, the function of muscles will be impaired. In cases of diffuse-type AVM, complete removal of lesions is almost impossible. If such a lesion is partially resected or only the feeding arteries are ligated, the remainder will soon regrow and the blood supplies recruit in large scale via collateral vessels. Subsequent attempted TAE via
arterial approach will be technically more difficult. In those unresectable AVMs, embolization may be the only choice in treatment [3].

With advance of high-speed high-quality digital imaging techniques, the feeding arteries and the extent of AVM can be well evaluated by angiography. TAE plays a more important role in AVM management than it used to do [4]. Vogelzang RL et al and Calligaro et al reported successful treatment of large pelvic AVMs by transcatheter embolization [5,6]. Some complications may be encountered after embolization, including extensive muscle necrosis, skin slough, nerve palsy and impotence due to non-target arteries embolization. Major risks of embolization are pulmonary embolization and reflux of particles to normal vascular structures.

To prevent these complications, superselective catheterization should be performed. The catheter should be placed near the nidus, which should be abolished as much as possible. If only feeding arteries are occluded, it will be very difficult to repeat embolization, like surgical ligations [7]. Multi-stages procedure should be planned especially for management of large AVM. In our experience, repeated embolotherapy is preferred at a 3-month interval when there is absent or minimal enlargement of collaterals.

Choosing appropriate embolization material is also very important. Variable embolization material such as gelantine sponge, PVA, absolute alcohol and tissue adhesive have been used with success in selected patients. Gelantine sponge is a kind of temporary embolization material, so it can not be used alone. Absolute alcohol can induce immediate thrombosis. It may lead to non-target embolization if the alcohol refluxes into normal vessels. Superselection and using the flow-directed micro-balloon catheter will reduce these complications. However, absolute alcohol is relatively tricky to use. Tissue adhesive is reported as a potential carcinogen and is not approved by the United States Food and Drug Administration. Furthermore, it is cumbersome to use clinically. So we chose PVA, a semi-permanent embolic agent, as the embolization material.

It is also quite important to choose particles of proper size. If the size of particles is too small, the risk of pulmonary embolization is increasing; but if the size is too large, the core nidus will not be completely abolished. So, we initially introduced the smaller size PVA and carefully monitored under fluoroscope. If the emboli passed through the AVM, the next larger size particles would be tried [8]. In case II, we initially infused PVA sized 150-250 µm. Series angiograms were obtained during the embolization procedure. When the blood flow that supplied the AVM slowed down, we injected larger PVA particles sized 355-500 µm to occlude the larger abnormal vessels. Finally Gelfoam pledgets were delivered to complete devascularization of arterial feeders to AVM after distal occlusion with PVA. Because Gelfoam is absorbable, reflux of small amount of pledgets into non-target vascular beds will do less harm to the tissue than PVA will [9]. In our cases, the AVMs were located in lower pelvis, and sexual function should be taken into consideration during embolization. Besides, both AVMs were large and supplied by multiple feeding arteries, so multi-stages embolization to reduce unexpected complications was planned and conducted.

Percutaneous sclerosis is another kind of therapy [10-13]. However, extralosomal ethanol extravasation may occur and result in necrosis of adjacent tissue. Under sonographic and fluoroscopic guidance, the incidence of this complication can be reduced [10,13]. Hence, embolization through intravascular catheter still provides greater safety and flexibility than direct puncture and permits wider range of embolizing material targeted for better obliteration of the core nidus. Percutaneous sclerosis of AVM should be tried only when intravascular catheter approach is technically difficult [11].

In conclusion, unresectable peripheral AVM can be managed with a dedicated angiographic unit and proper selection of embolic materials on a multi-stages basis. The treatment of AVM should be aimed at permanent obliteration of the core nidus and avoidance of inadvertent non-target embolization that results in tissue necrosis and nerve injuries.

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大型週邊動靜脈畸型經動脈血管栓塞術：
二個病例報告

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周邊先天性動靜脈畸型的治療對臨床醫師是一大挑戰，在無法以手術完全切除的動靜脈畸型，血管栓塞術是另一種選擇。我們報告兩個在下肢的大型動靜脈畸型病例，開刀後發現無法完全切除且傷口持續滄血及癒合不良，嘗試經動脈血管栓塞術治療，其症狀皆有大幅改善，而且並沒有明顯的合併症出現。

關鍵詞：動靜脈畸型；四肢，經動脈血管栓塞術