



# Successful Endovascular Treatment of a Ruptured Giant Lumbar Arteriovenous Malformation

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Arteriovenous malformations (AVMs) are rare congenital vascular anomalies where rupture can be fatal. This report describes a case of a 69-year-old woman who presented with sudden severe lower back pain radiating to her left buttock and thigh and an inability to move her left leg. Computed tomography angiography (CTA) showed an extensive ruptured retroperitoneal AVM on the left paravertebral side with active bleeding and a large hematoma. Cardiopulmonary resuscitation with considerable blood transfusion was required. The AVM was effectively treated with coil embolization of the feeding lumbar arteries. Shortly after the intervention, the patient developed abdominal compartment syndrome. Urgent laparotomy with evacuation of the retroperitoneal hematoma was required. The patient was discharged 16 days after admission in a good physical condition and has been free of symptoms for more than a year after intervention. However, follow-up CTA showed residual contrast enhancement in the AVM and further surveillance is required.

**Key Words:** Vascular malformations, Arteriovenous malformations, Lumbar artery, Ruptured aneurysm, Therapeutic embolization

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## INTRODUCTION

Vascular malformations (VMs) are congenital structural anomalies that are often detected at birth [1,2]. However, some VMs manifest later in childhood or even adulthood [2]. According to the current understanding of the biology and genetics of VMs, the International Society for the Study of Vascular Anomalies (ISSVA) classified VMs into the following 4 categories: A) simple VMs; B) combined VMs (defined as 2 or more types of vascular anomalies found in 1 lesion); C) VMs of major named vessels; and D) VMs associated with other anomalies [1]. From a clinical perspective, VMs may be classified based on hemodynamic findings into slow-flow, fast-flow, or combined malformations [2]. Arteriovenous malformations (AVMs) are fast-flow malformations characterized by abnormal connections between arteries and veins without an interposition of capillary vessels [1,2].

The most common location for AVMs is intracranial, followed by the limbs, trunk, and viscera [2]. AVM expansion can lead to functional impairment due to local destruction, ischemia due to steal phenomenon, high output cardiac failure, or bleeding due to rupture. So far, little is known about factors that trigger expansion, however, it often seems associated with pregnancy, puberty, or secondary to local trauma [1]. Vascular specialists usually recommend treatment of symptomatic AVMs, however, available evidence is scarce and currently no treatment guidelines exist.

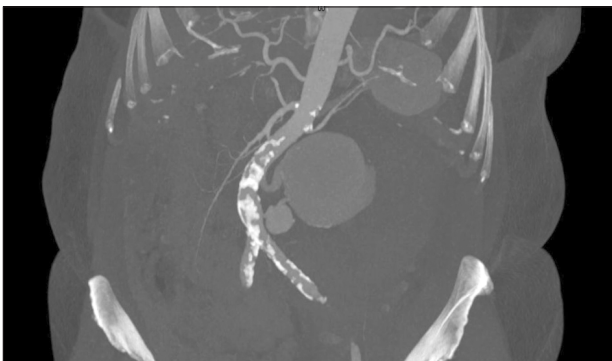
Lee et al. [3] presented a series of 76 patients with AVMs managed by a multidisciplinary team, including both endovascular and surgical treatment options. The results at 24 months were optimal for surgically accessible lesions with staged management, including preoperative embolization or sclerotherapy and consecutive surgical resection. The results for surgically inaccessible lesions treated with an

endovascular approach were only slightly inferior showing satisfactory outcomes in 25/32 patients and good to fair results for the remaining 7 patients.

This report describes a rare case of a patient with a ruptured giant lumbar AVM requiring emergent intervention with resuscitation. Informed consent was obtained from the patient to present her case in anonymized form to experts in the field.

## CASE

A 69-year-old woman was referred to the emergency department due to sudden severe lower back pain with radiation to her left buttock and thigh as well as an inability to move her left leg. Initially, radicular irritation was suspected as a physical examination revealed hypoesthesia at the left anterolateral thigh and reduced strength of the left hip flexors (M4, British Medical Research Council Muscle Scale) [4]. The abdomen was soft with a large indolent umbilical hernia and bilateral foot pulses were palpable. During the clinical examination, the patient had a short syncope with spontaneous recovery after few seconds. The patient's blood pressure was measured shortly after this syncope and she was normotensive. A vasovagal reaction was suspected. Subsequent ultrasonography showed an extensive aneurysmal vessel in the lower abdomen. The patient was suspected to have a ruptured aortoiliac aneurysm. Initial hemoglobin level was 8.7 g/dL (normal range, 12.0 to 16.0 g/dL) showing anemia. An emergency computed tomography angiography (CTA) showed multiple ectatic and elongated lumbar arteries on the left paravertebral side (L2 to L4) leading into draining varicose veins in the gluteal subcutaneous tissue on the left side (Supplementary Video). The lumbar artery L3 was aneurysmal with a maximum diameter of 8.2 cm. The aneurysm was ruptured with active bleeding and



**Fig. 1.** Reconstructed initial computed tomography angiography showing a retroperitoneal arteriovenous malformation on the left paravertebral side with ectatic feeding lumbar arteries.

hematoma displacing the left kidney and compressing the descending colon (Fig. 1). In summary, a ruptured lumbar AVM with active bleeding and extensive retroperitoneal hematoma was diagnosed. The aorta and iliac vessels were normal sized and the umbilical hernia showed no signs of incarceration.

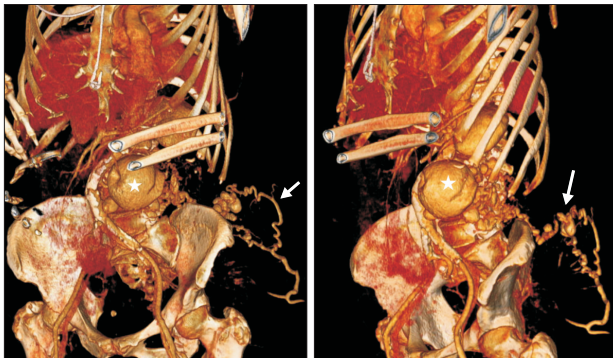
Urgent endovascular coiling of the feeding lumbar arteries was proposed; however, the patient experienced hemodynamic collapse (a drop in systolic blood pressure from 120 to 70 mm Hg) and lost consciousness. Immediate airway intubation and a major blood transfusion (10 bags of packed red blood cells, 3 bags of fresh frozen plasma, 500 IE prothrombin complex concentrate, and 2,500 IE factor XIII) were required to stabilize the circulation. At the same time, arterial access was established at the right common femoral artery (6 Fr introducer sheath). Aortic angiography using a 4 F OmniFlush (AngioDynamics Inc., Queensbury, NY, USA) showed dilated lumbar arteries at segments L2 to L4 on the left side, all feeding the ruptured aneurysm of the L3 artery (diameter, 8.2 cm). In addition, arterial-feeding branches came from the left internal and external iliac arteries (Fig. 2, 3). Stepwise cannulation using a 5 F SOS end-hole catheter (AngioDynamics Inc.) was performed to occlude the 3 main lumbar feeding vessels using detachable embolization coils (3- and 4-mm, 2D-Helical; Boston Scientific, Marlborough, MA, USA). Completion angiography showed reduced and delayed residual contrast enhancement of the AVM via backdoor feeding from a left gluteal vein. Active bleeding was no longer detected and the patient became hemodynamically stable.

At the intensive care unit, an intra-abdominal pressure (i.e., intravesical pressure) of 24 mm Hg was measured (normal range, 5 to 7 mm Hg). Due to the abdominal compartment syndrome, the umbilical hernia was incarcerated. Urgent laparotomy was performed. The hernia contained parts of the transverse colon, multiple jejunal loops, and parts of the omentum; however, no signs of tissue necrosis were present. The retroperitoneal hematoma had ruptured into the abdominal cavity, but upon evacuation no active bleeding was detected. The abdomen was closed using a Symbotex Composite Mesh (Medtronic, Minneapolis, MN, USA). To avoid recurrent abdominal compartment syndrome, the mesh was placed between the edges of the fascia (inlay bridging technique) thereby extending the abdominal cavity.

The patient then recovered quickly. The left sided lumbar radicular pain decreased but remained to some degree, while the paresis had resolved completely. The varicose veins in her left buttocks remained. CTA after 1 week showed no residual perfusion of the AVM. The patient was discharged at postoperative day 16 in good physical condi-

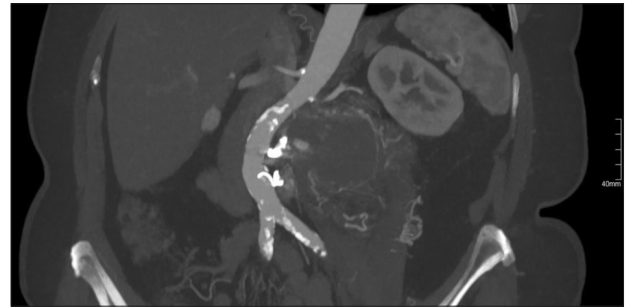


**Fig. 2.** Digital subtraction angiography showing dilated lumbar arteries at segments L2 to L4, feeding a giant lumbar arteriovenous malformation with active bleeding, mainly from L3.

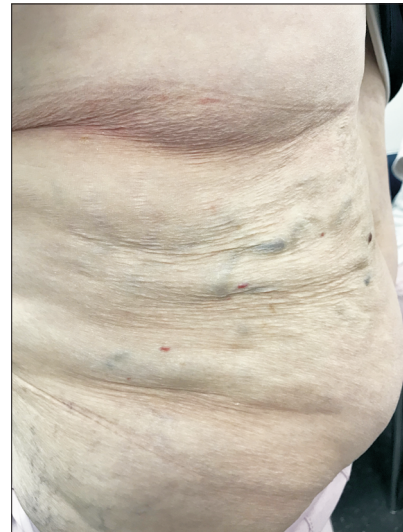


**Fig. 3.** Three-dimensionally reconstructed computed tomography angiography prior to intervention showing the lumbar arteriovenous malformation (asterisks) with ectatic lumbar arteries and draining varicose gluteal veins (arrows).

tion. At 4 months postoperative, a routine follow-up CTA was performed. The AVM had slightly increased in size and perfusion had reappeared, yet without active bleeding. Three residual feeders (L2 and 3 left, and 1 external iliac artery feeder) were coiled in 2 separate sessions. The final angiography showed low-flow residual perfusion of the AVM that was no longer accessible endovascularly due to packed coils. Two further CTAs were performed after 10 and 15 months. Both showed a stable size of AVM with minimal regression of the retroperitoneal hematoma but a slight increase in the contrast enhancement in the AVM (Fig. 4) and the varicose veins in her left buttock (Fig. 5). Although a remaining and slightly increasing perfusion of



**Fig. 4.** Reconstructed post-interventional computed tomography angiography after 15 months showing the stable size of the arteriovenous malformation (AVM) with residual contrast enhancement in the AVM and coils at lumbar level L2 and L3.



**Fig. 5.** Dorsolateral view of the patient showing persisting varicose veins at the lower left back, 1 year after initial treatment.

the AVM is present, the patient has remained asymptomatic to date.

## DISCUSSION

Here, we presented an uncommon case of a patient with a ruptured giant lumbar AVM, an extremely rare vascular pathology where the exact etiology is not well understood. Correct identification of this vascular anomaly was essential, and appropriate imaging was a prerequisite for appropriate management.

AVMs do not follow the typical arterial tree structure, but several arterial vessels can feed the AVM with consecutive arterialization and high-pressure blood flow into the venous system [2]. If an AVM ruptures, the surrounding

connective tissue may limit the bleeding initially. In this case, the retroperitoneal tissue tamponaded the bleeding. Surgical dissection of the surrounding tissue can be detrimental in this situation due to uncontrollable bleeding once the natural tamponade is removed. Further, lessons learned from the management of ruptured abdominal aortic aneurysms can be applied in this situation. The concept of permissive arterial hypotension with delayed volume resuscitation may be lifesaving. Volume resuscitation should be delayed until bleeding control is achieved as long as the patient is conscious [5]. Hence, fast and correct identification of the pathology is paramount for appropriate interdisciplinary management.

Embolic agents have been used since the 1980s to treat VMs [6]. Today, a wide armamentarium for endovascular therapy of VM is available. Endovascular approaches can be divided into solid embolic tools that directly induce thrombosis (e.g., particles, plugs, coils, or detachable balloons) or sclerosant liquid agents that destroy the endothelial cells and thereby induce thrombosis indirectly (e.g., bleomycin, ethanol, sodium tetradecyl sulphate, or others) [7-9]. The advantage of solid embolic tools is the possibility of controlled placement and potential immediate hemostatic control due to occlusion. However, ischemic endothelial cells (due to thrombosis after embolization) may excrete angiogenic factors stimulating neovascularization [10]. As a consequence, neovascularization is frequently observed in post-embolization imaging [10]. In contrast, sclerosant liquid agents destroy the endothelial cells and address this problem at a cellular level [10]. However, major complications can be provoked by liquid agents including deep vein thrombosis, pulmonary embolization, or local injuries [10,11].

Currently, there are no practice guidelines for the periprocedural management of endovascular treated AVMs. In an emergency situation, rapid hemostasis is essential as the primary treatment objective. In this case, initial coiling proved an effective and life-saving strategy in the emergency situation. However, as in an endovascular repair of a ruptured aortic aneurysm, continued bleeding may result in abdominal compartment syndrome, a potentially deleterious condition [12]. Hence, close monitoring of the intra-abdominal pressure and surgical back-up, in case rapid decompression is needed, must be part of the interdisciplinary management of patients with ruptured AVMs.

Due to the reticular vascular structure of AVMs with increased risk of neovascularization after local thrombosis, surveillance is necessary even in cases without any residual AVM perfusion. In this case, we will continue surveillance despite the current steady-state situation. In case of further growth, local symptoms, or recurring high flow measured by ultrasound in the varicose veins, preemptive repeat coil/

glue embolization will be performed. In this instance we would prefer a percutaneous direct puncture of the AVM since the endovascular cannulation was technically unfeasible in the last attempt. A surgical approach should probably still be avoided as decompression of the organized surrounding tissue may eventually lead to uncontrollable bleeding.

In conclusion, VMs require interdisciplinary and individually tailored management. In this 69-year-old female patient, staged management for a life-threatening retroperitoneal AVM rupture with emergent endovascular coil embolization and subsequent surgical decompression of an abdominal compartment syndrome was a sustainable treatment approach, but surveillance will have to continue.

## SUPPLEMENTARY MATERIAL

Supplementary data can be found via <https://doi.org/10.5758/vsi.2020.36.1.33>.

## CONFLICTS OF INTEREST

The authors have nothing to disclose.

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Concept and design: LM, NG, FD. Analysis and interpretation: LM, NG, FD. Data collection: LM. Writing the article: LM. Critical revision of the article: NG, FD. Final approval of the article: LM, NG, FD. Obtained funding: none. Overall responsibility: FD.



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