

Case Report

Thoraco-lumbar artery aneurysms associated with a metameric paraspinal lesion presenting with retroperitoneal hemorrhage: Endovascular management

Alejandro Santillan, Walter Zink, Athos Patsalides, Y. Pierre Gobin

Division of Interventional Neuroradiology, Department of Neurological Surgery, New York Presbyterian Hospital, Weill Cornell Medical Center, New York, NY, USA

E-mail: *Alejandro Santillan - als2052@med.cornell.edu; Walter Zink - waz2002@nyp.org; Athos Patsalides - atp9002@med.cornell.edu;

Y. Pierre Gobin - yvg2001@med.cornell.edu

*Corresponding author

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Abstract

Background: Retroperitoneal hemorrhage is a life-threatening condition. This is the first reported case of rupture of one of multiple thoraco-lumbar artery aneurysms associated with a metameric paraspinal vascular lesion.

Case Description: A 77-year-old female patient presented to the emergency room with a new onset of left-sided low back pain shooting down the leg associated with weakness, numbness, and inability to walk. On physical examination, there was a notable left paraspinal swelling with a harsh bruit audible in the same area, left flank ecchymosis and a positive straight leg raising test. A computed tomography (CT) scan showed a large retroperitoneal hematoma. Digital subtraction angiography showed a large left paraspinal high-flow arteriovenous lesion, with large arterial aneurysms of the left T11, T12, and L1 segmental arteries. The patient was successfully treated with endovascular aneurysm embolization using coils and Onyx-34. Six months following the procedure, the patient had fully recovered, and a follow-up angiogram showed no residual or recurrent aneurysms.

Conclusion: Thoraco-lumbar artery aneurysms have never previously been described in association with a metameric paraspinal vascular malformation. We report a case of retroperitoneal hemorrhage due to rupture of one of several high-flow artery aneurysms of a paraspinal arteriovenous malformation (AVM). The diagnosis was made on CTA, MRI, and angiography, and the lesion was successfully treated by transarterial embolization.

Key Words: Arteriovenous malformation, endovascular embolization, metameric lesion, Onyx-34

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INTRODUCTION

Metameric paraspinal AVMs are exceptional and there are only a few reported cases in the literature.^[14,17] It is believed that these lesions are congenital, extramedullary,

extradural and can involve bone, soft tissue, and muscle.^[13]

We report a rare case of a metameric paraspinal AVM associated with high-flow artery aneurysms presenting with retroperitoneal hemorrhage.

CASE REPORT

A 77-year-old Cantonese speaking female patient with a vague history of “back tumor” 15 years ago that was never biopsied, presented to the emergency room with a new onset of left-sided low back pain shooting down the leg associated with weakness, numbness, and inability to walk. She was also complaining of nausea and dizziness. The patient was hemodynamically stable (blood pressure 125/65 mmHg; pulse rate, 70/min) although her work-up revealed a hemoglobin of 7.8 g/dL. On physical examination, there was a notable left paraspinal swelling with a harsh bruit audible in the same area associated with tenderness in the thoracolumbar junction, left flank ecchymosis and a positive straight leg raising test. The patellar and Achilles tendon reflexes were normal and symmetric. There was no sensory disturbance. With a consideration for internal bleeding, fluid resuscitation, blood transfusion, and oxygen supplementation were administered. The patient received transfusion of 4 units of packed red blood cells and 1 unit of platelets. A computed tomography (CT) scan showed a large retroperitoneal hematoma [Figure 1a], and computed tomography angiography (CTA) revealed several large, dilated vascular structures in the left retroperitoneum [Figure 2a]. Magnetic resonance imaging (MRI) of the spine confirmed a large hematoma in the left psoas muscle extending from the T12 to L4 vertebral levels and numerous flow-voids in the left paraspinal muscles [Figures 1b and 2b]. No abnormal signal of the spinal cord was noted.

The patient was taken urgently to the angiography suite. From a right femoral artery access, a spinal angiography

was performed. It demonstrated a large left paraspinal high-flow arteriovenous malformation (AVM), with large arterial aneurysms at the left T11, T12, and L1 segmental arteries [Figure 3]. The segmental arteries, and then the aneurysms were selectively catheterized and embolized. The left L1 aneurysm was embolized using three Axium coils (ev3; Irvine, CA), followed by injection of Onyx-34 (Micro Therapeutics Inc.; Irvine, CA). The left T11 and T12 aneurysms were embolized with intra-aneurysmal injection of Onyx-34 [Figure 4]. The postoperative course was uneventful, and there was no recurrence of hemorrhage. The patient was seen for clinical follow-up 1 month after treatment and had no residual back or leg pain. Six months following the procedure, the patient underwent an MRI [Figure 5] and a conventional spinal angiography [Figure 6]. The angiogram showed no residual or recurrent aneurysms, but persistence of the AVM in the left T11-L1 paraspinal region. We decided not to perform embolization of the residual AVM itself because we estimated that the risk of hemorrhage was low as long as the aneurysms were occluded. The patient will be followed with yearly MRIs.

DISCUSSION

Paraspinal AVMs are more commonly diagnosed in a pediatric population,^[11] and are often located in the thoracic and cervical regions.^[5,11] The AVM can involve the paravertebral musculature, nerve root foramina, prevertebral region, and sometimes enter the spinal canal, and may have paraspinal and epidural venous drainage.^[5,7,11,15,17] In the case reported here, the AVM presented with a strict paraspinal venous drainage without

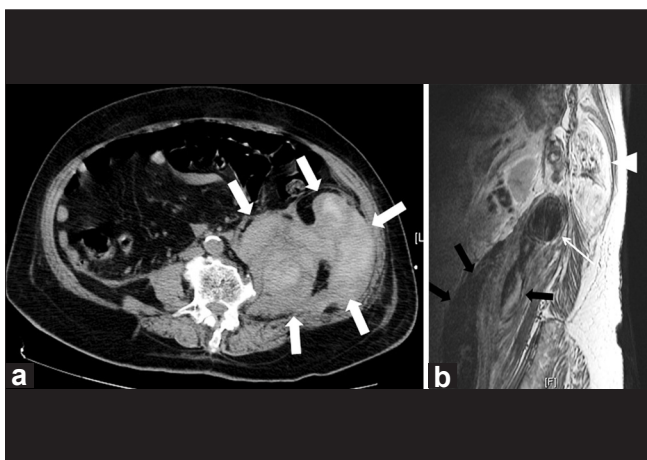


Figure 1: (a) A non-contrast axial CT scan shows a large left-sided retroperitoneal hematoma (arrows) extending from T12 to L4 levels, and measuring 9.2 x 6.8 cm. (b) A sagittal T2-weighted MR image shows the paraspinal vascular lesion (arrowhead), and a hematoma in and around the left psoas muscle (black arrows) resulting from rupture of the left L1 feeding-artery aneurysm (thin arrow)

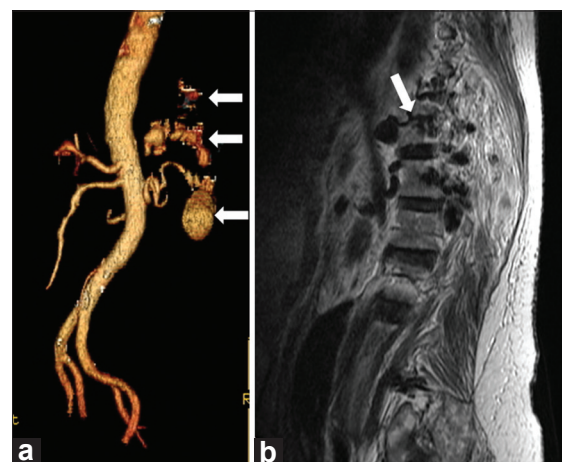


Figure 2: (a) A 3D CT angiography reveals large aneurysms originating from the left T11, T12, and L1 segmental arteries (arrows). (b) Sagittal T2-weighted MR showing a paraspinal vascular malformation with multiple flow-voids, and a direct communication (arrow) between the malformation and a T12 high-flow artery aneurysm



Figure 3: Digital subtraction angiography of the left segmental arteries T11 (a), T12 (b), and L1 (c) showing large high-flow aneurysms

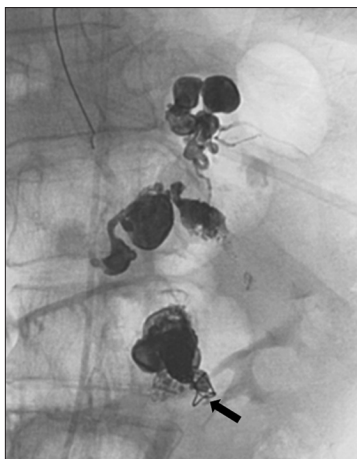


Figure 4: Post-embolization angiogram (unsubtracted view) showing the mass of coils in the L1 segmental artery (arrow), and Onyx-34 in the aneurysms originated from T11, T12, and L1 segmental arteries

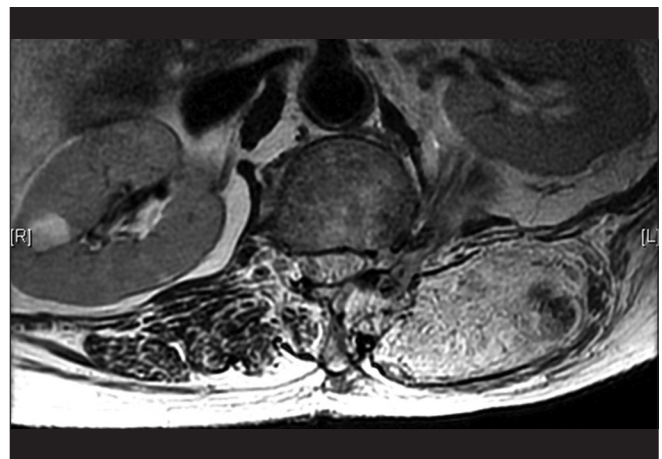


Figure 5: MR images obtained at 6 months after embolization. Axial T2-weighted MR image showing increased signal on T2 in the left paraspinous region from numerous flow-voids largely resolved secondary to embolization



Figure 6: Digital subtraction angiogram 6 months following embolization, showing a residual vascular malformation in the paraspinous muscles with a feeding artery coming off the left T11 segmental artery

involvement of the epidural venous plexus. To the best of the authors' knowledge, this is the first reported case of a paraspinous AVM presenting with retroperitoneal hemorrhage.

In our patient, the paraspinous AVM had three large aneurysms arising from T11, T12, and L1 segmental arteries. The large hematoma located in the left psoas muscle, extending from T12 to L4 levels, probably occurred from rupture of one of these aneurysms, most likely the L1 segmental artery aneurysm since it was the largest, and had a very irregular shape. It is important to notice that there were no cutaneous vascular lesions in our patient, as if the lesion would have extended into the skin with its corresponding dermatomes; we could have diagnosed a Cobb syndrome.^[8,9]

Retroperitoneal hemorrhage is a rare condition which

requires a high index of clinical suspicion. It may occur as a complication of femoral angiography, ruptured abdominal aortic, iliac, renal, mesenteric, and ovarian artery aneurysms,^[2,3,12] or spontaneously in patients who are anticoagulated or on hemodialysis.^[1,6] Preoperative imaging (including urgent CT scanning, MR imaging, and spinal angiography) is indicated in the setting of a retroperitoneal hemorrhage with or without hemodynamic instability.^[1,4,6]

The indications for treatment of paraspinal AVMs is usually based on common clinical findings, such as back pain, paravertebral murmur, high-output cardiac failure, paraparesis, or paraplegia (due to direct compression, steal phenomenon, intraspinal hemorrhage, or venous hypertension), dysphagia, and spinal instability with or without major spinal deformity.^[9,11,17] In the case reported, the patient presented to the emergency room with different symptomatology in the setting of retroperitoneal bleeding (anemia, flank ecchymosis, back pain, and a positive straight leg raising test). Since the lesion did not invade the epidural space or spinal canal, our patient had no symptoms of spinal cord dysfunction.

Endovascular management is an efficient treatment option for paraspinal AVMs.^[9,13,14,16] Different embolic materials have been used for the treatment of these lesions like balloons, particles, NBCA, platinum coils,^[10,13,17] and recently Onyx.^[11] In our case, we used coils and Onyx-34 since our primary goal was to embolize high-flow aneurysms. Embolization with permanent occlusive agents is an effective way to treat these rare but potentially life-threatening lesions.

CONCLUSION

We present a unique case of an adult patient with thoraco-lumbar artery aneurysms associated with a metameric paraspinal arteriovenous malformation presenting with retroperitoneal hemorrhage due to rupture of one of multiple aneurysms. The lesion was diagnosed with CTA, MRI, and angiography, and successfully treated by endovascular embolization using coils and Onyx-34. Transarterial embolization was the therapeutic approach of choice in the reported case provided the patient was hemodynamically stable. Observation of the residual vascular malformation will

be necessary because additional embolization or surgical treatment might be needed by the patient in the future.

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