

## Embolization followed by surgery for treatment of perimedullary arteriovenous fistula causing acute myelopathy

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### Abstract

**Background:** Perimedullary arteriovenous fistula (AVF) is rare. There are three subtypes, and the treatment strategies for each are different. Subtype B (multiple fistulas) can be treated by either embolization or surgery. On the basis of a case from our treatment experience, we propose a method for achieving optimal outcome while minimizing nerve injury.

**Case Description:** A 51-year-old female was admitted to our hospital with acute myelopathy caused by a perimedullary AVF. Initially, we treated her by embolization using the chemical agent Onyx. Her symptoms improved immediately but gradually returned beginning 1 week later. Two months later, the symptoms had returned to pretreatment status, so we removed the fistulas surgically. Severe adhesions between nerve and occult venous varices were noted during the operation. Afterward, the patient's symptoms improved significantly. Histopathological sections showed an inflammatory reaction around the varices.

**Conclusions:** We initially considered several possible reasons for the return of symptoms: (a) Hypoperfusion of the spinal cord; (b) mass effect of the occult vein varices; (c) residual AVF or vascular remodeling resulting in recurrent cord hypertension; (d) Onyx-induced perivascular inflammation resulting in nerves adhering to each other and to occult venous varices. Clinical, surgical, and pathological findings ruled out the first three, leaving Onyx-induced perivascular inflammation as the probable reason. Given our treatment experience and the pros and cons of the two methods, we propose that initial embolization followed by surgery after 5 days to remove occult venous varices is the ideal strategy for treating perimedullary AVF of subtype B.

**Key Words:** Embolic material, Onyx, perimedullary arteriovenous fistula, spine

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## INTRODUCTION

Spinal vascular malformations are rare and can generally be divided into four types:<sup>[2]</sup> Spinal dural arteriovenous fistula (AVF) (type I), glomus-type intramedullary

arteriovenous malformations (type II), juvenile-type intramedullary arteriovenous malformations (type III), and perimedullary AVF (type IV). Type IV has three subtypes, classified according to size and the number of feeding arteries and drainage veins. Subtype A is a single

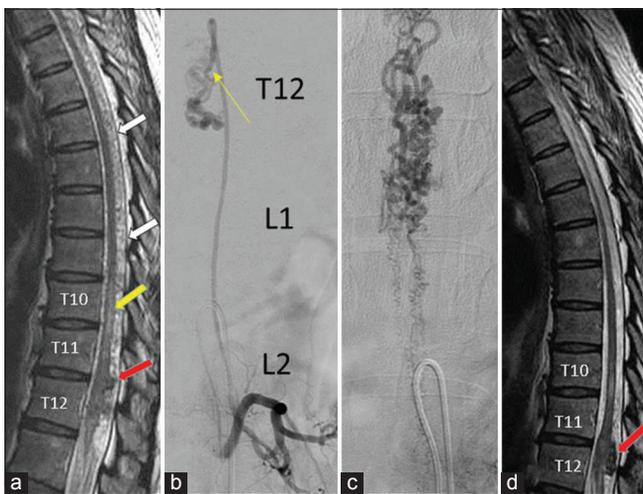
small fistula fed by a single small artery; it is best treated through surgical resection, as the small size of the feeding artery can make microcatheter embolization difficult. Subtype B is defined as multiple medium fistulas fed by multiple arteries; Gueguen *et al.* report that it can be treated by surgery or embolization.<sup>[4]</sup> Subtype C is a giant fistula with multiple large, dilated arterial feeders and is best treated by embolization. The best treatment strategy for subtype B is still being debated. We report the case of a 51-year-old female who suffered from progressive myelopathy and harbored an intradural perimedullary AVF that caused venous hypertension and compression of the conus medullaris and cauda equina. Transarterial embolization and surgical resection were performed, and 6 months after surgery, the patient had completely recovered except for slight residual leg numbness.

### CASE DESCRIPTION

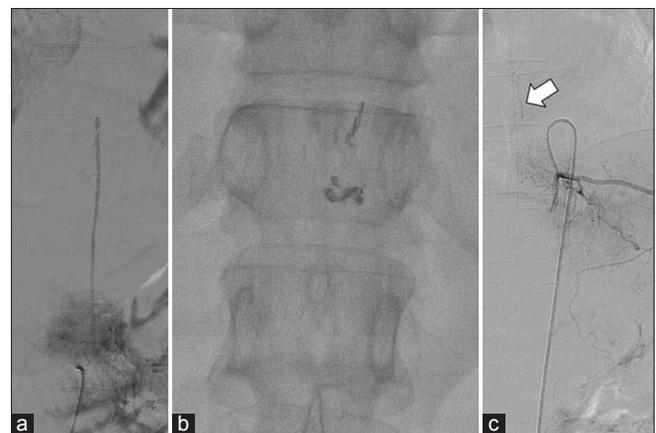
A 51-year-old female was admitted for progressive ascending spastic paraparesis and hypesthesia of 6 months' duration extending from her feet to her buttocks. Neurologic examination revealed that light-touch sensation below T10 was impaired and that muscle strength was Medical Research Council grade 4 in both lower limbs. Additional myelopathy symptoms included bilateral extensor plantar response, intensified deep tendon reflexes in both knees and Achilles tendons, and fecal and urinary incontinence. A thoracic spinal lesion was suspected. Magnetic resonance imaging (MRI) was performed on the thoracic spine and showed a perimedullary vascular lesion [Figure 1]. Subsequent

angiography showed a major feeding artery from the left L2 lumbar artery and another minor feeding artery from L1. Multiple fistulas drained by multiple perimedullary veins were present at the T12 level [Figure 1], indicating a typical spinal vascular malformation of type IV, subtype B. After discussing her treatment choices with us, the patient chose transarterial embolization, which was performed by injecting the chemical agent Onyx (Covidien, Plymouth, MN, USA) via a coaxial microcatheter system. An angiogram taken, a transarterial embolization, showed complete occlusion of the fistulas with preservation of the parent artery [Figure 2].

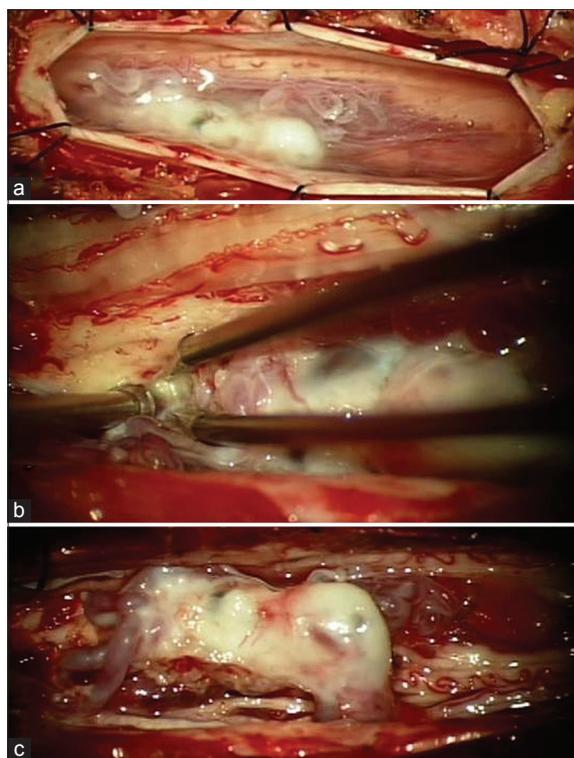
The symptoms improved immediately, and only residual foot numbness remained. To prevent spinal cord swelling, dexamethasone (4 mg every 8 h) was administered by intravenous drip for 1 week. Unfortunately, myelopathy symptoms came back gradually beginning 1 week after embolization and had returned to their pretreatment status 2 months later. Accordingly, we suggested follow-up catheter angiography to screen for vascular remodeling, but the patient refused it. Instead, we performed thoracic spine MRI to rule out the possibility that residual AV shunting was causing recurrent venous hypertension and cord hypoperfusion. The lesion still showed vascularity and edema, although more less than on the previous MRI [Figure 1]. As the patient's myelopathy has persisted, we decided to remove this lesion immediately through open surgery; a posterior approach at the level of T11 and T12 was used to remove it, and then laminoplasty was performed. The operation revealed a mass of occult, white venous varices floating in the cerebrospinal fluid that had formed severe adhesions to surrounding nerves. We carefully lysed the adhesion then dissected the lesion from the nerves with a nonstick bipolar forceps and microscissors and totally removed it [Figure 3]. During the lysis procedure, we found no red veins, bleeding, or oozing, or other signs



**Figure 1:** (a) T2WI MRI showing a hypointense lesion (red arrow) behind the spinal cord at T11 and T12 with peripheral tube-like lesions (white arrow), fistulas, and perimedullary venous drainage. At the T10 level (yellow arrow), it shows a hyperintense signal, indicating cord edema related to venous hypertension. (b) Angiogram showing the major feeding artery from the left L2 lumbar artery and a fistula at the T12 level. (c) Venous phase showing perimedullary venous drainage. (d) MRI taken after surgery showing decreased lesion size, cord edema, and vascularity compared with the previous MRI



**Figure 2:** (a and b) The arteriovenous fistula was completely occluded after embolization. (c) Postembolization angiogram of the left artery of Adamkiewicz (white arrow) at the T10 level. Note, no residual fistula and no occlusion of the anterior spinal artery



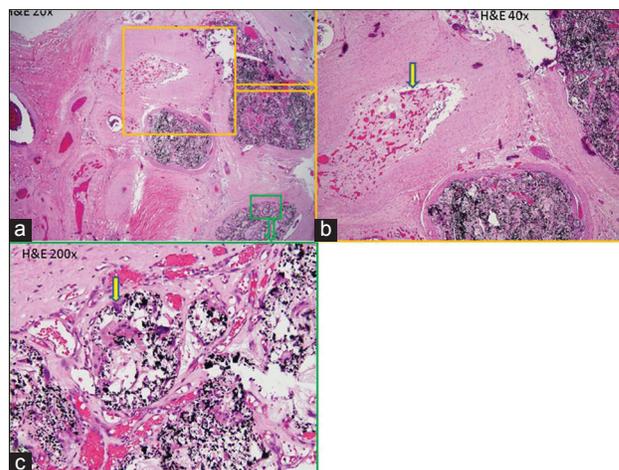
**Figure 3: (a) The operation revealed occult white venous varices floating in CSF. (b) The varices formed severe adhesions with the surrounding nerves. (c) The dissected occult white venous varices and no red vein were noted**

of residual arteriovenous shunting, and optimal blood loss was minimal. Histopathological sections showed an inflammatory reaction around the varices [Figure 4].

After the operation, the patient's muscle strength returned to normal, her incontinence disappeared, and her sensation almost completely recovered. We followed up this patient for 6 months and noted no further symptoms except mild residual left leg numbness.

## DISCUSSION

In patients with a perimedullary AVF, venous hypertension is the main cause of neurological dysfunction.<sup>[4]</sup> Surgical removal is the most direct treatment method but may lead to massive bleeding and require prolonged follow-up of angioarchitecture.<sup>[5]</sup> Embolization is minimally invasive, but the potential for recanalization must be considered. Further trials are needed to determine the best treatment.<sup>[3]</sup> Cho *et al.*<sup>[1]</sup> reported that surgery alone achieved a better outcome than embolization alone in a group of patients with type IV-B AVF. In our case, initial transarterial embolization successfully occluded the fistulas and decreased venous hypertension [Figure 1]. The myelopathy symptoms improved immediately but returned 1 week later. We initially considered four possible reasons for this occurrence: (a) Hypoperfusion of the spinal cord; (b) mass effect of the occult venous



**Figure 4: (a and c) Photomicrographs of vessels embolized with Onyx showing giant cells forming as a foreign-body reaction (yellow arrows). Hematoxylin–eosin staining, original magnification  $\times 20$  (a) and  $\times 200$  (c). (b) The Onyx cast lined by endothelial cells**

varices; (c) residual AVF or vascular remodeling resulted in cord hypertension again; and (d) Onyx-induced perivascular inflammation and resulting in nerves adhering to each other and to occult venous varices. Clinical examination, postembolization MRI, and the findings of the operation ruled out the first three.

According to Natarajan *et al.*,<sup>[6]</sup> the use of Onyx in embolization may result in vascular or perivascular inflammation, penetration of the embolic agent, or angionecrosis. They observed that  $>8$  days after Onyx embolization, giant cells formed as a foreign-body reaction in 12 of 22 patients; this may explain the timing of the return of symptoms in our case after dexamethasone was discontinued.

On the basis of our treatment experience, we propose that initial embolization, followed by surgery 5 days later to remove occult varices, is the best treatment strategy for AVF type IV-B AVF. Performing embolization first can decrease operation blood loss and duration. If significant vascularity remains after embolization, antiplatelet drugs should be administered to prevent excessive venous thrombosis. Steroid therapy and surgery must be implemented as soon as possible after embolization to forestall potential chemical injury to nerves, secondary mass effect, and neovascular generation.

## Conflicts of Interest/Disclosures

The authors declare that they have no financial or other conflicts of interest in relation to this research and its publication.

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