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Case Report

Hybrid open-endovascular onyx embolization of spinal type IVb perimedullary spinal arteriovenous fistula through direct posterior spinal vein access: A case report

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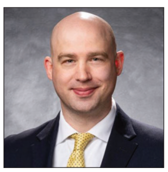

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ABSTRACT

Background: Spinal arteriovenous fistulas (SAVFs) are direct communication between arteries and veins without intervening abnormal vessel nidus, which often results in venous congestion and spinal cord dysfunction. Ventrally located SAVF can be challenging to treat through traditional open or endovascular approaches.

Case Description: We describe a hybrid (open/endovascular) procedure in a 72-year-old male with a Takai Type IVb SAVF presenting with paraparesis and sphincter dysfunction. Imaging revealed a conus medullaris SAVF in which the main fistulous connection was located ventrally. The conventional endovascular approach was deemed risky, and open surgery failed in the first attempt. The SAVF was resolved using a hybrid approach: under direct visualization, an engorged dorsal vein was punctured with an Angiocath, and a fluoroscopy-guided microcatheter was advanced through it to reach and embolize the ventral perimedullary fistulous connection. After surgery, his progressive neurological decline stabilized, radiographic spinal cord edema improved, and follow-up angiography confirmed obliteration of the fistula. Neurological function remained at the preoperative baseline.

Conclusion: This approach may be a treatment for selected cases of type IVb SAVF. Easily accessible feeding vessels are coagulated and cut; the inaccessible ones can be embolized endovascularly during the same procedure.

Keywords: Case report, Embolization, Endovascular, Hybrid, Spinal arteriovenous fistula, Spine

INTRODUCTION

Spinal arteriovenous fistulas (SAVFs) are direct communication between arteries and veins without intervening abnormal vessel nidus, which often results in venous congestion and spinal cord dysfunction.[12] Occlusion of these fistulas is associated with stabilization or reversal of neurological dysfunction.[5] SAVF is included in numerous classification schemes for spinal arteriovenous malformations/arteriovenous shunts, such as those described by Di chiro *et al.*, Rosenblum *et al.*, Mourier *et al.*, Spetzler *et al.*, Rodesch *et al.*, Geibprasert *et al.*, and Rangel-Castilla *et al.*, and Takai.[1,3,4,6,7,9,11,13,14,16,17] The proposed classification of spinal arteriovenous

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shunts by Takai describes three types of SAVF: Type I (dural arteriovenous fistula), Type IV (perimedullary arteriovenous fistula), and Type V (extradural arteriovenous fistula). Takai's classification also divides type IV SAVF into Type IVa (single feeder), IVb (multiple feeders/medium size), and Type IVc (multiple feeders/giant size). Type IVb and IVc are particularly challenging given the multiple feeding vessels and frequent ventral points of fistulization. Open microsurgical ligation, endovascular, and combined open and endovascular approaches have all been described to approach type IV SAVF. The decision for endovascular or open treatment is dependent on the lesion's hemodynamics, location, and angioarchitecture.[10] While reaching ventral SAVFs located above the conus medullaris through open surgery is challenging, the small diameter of the distal anterior spinal artery and the risk of spinal cord ischemia are obstacles to the endovascular transarterial approach. Similarly, the transvenous route faces obstacles, including long access distances and vessel tortuosity. Although the hybrid management of SAVFs has been previously described,[8,18] here we describe a novel closure using liquid embolic through a combined open/endovascular approach with transvenous catheter access through puncture of a perimedullary draining vein of a Type IVb SAVF.

CLINICAL PRESENTATION

A 72-year-old man presented with a history of hypertension, cirrhosis (Child-Pugh A), pituitary macroadenoma, status post-endoscopic endonasal resection complicated by postoperative hemorrhage, and multiple failed attempts at lumbar drain placement who had progressive gait ataxia, back pain, urinary retention, and headaches. Spine magnetic resonance imaging revealed a low-lying conus ending at L3–4, multiple intradural cysts suggestive of arachnoid adhesions, and abnormal vessels suggestive of a SAVF [Figure 1a]. Spinal angiogram [Figures 1b and c] confirmed a type IV SAVF.

Given progressive neurological symptoms, treatment was recommended. Endovascular treatment was not attempted due to the unclear location of the fistulous point and the high risk of spinal cord ischemia from direct anterior spinal artery catheterization. Surgical treatment was undertaken with laminectomies from T12 to L2 as possible fistula connections were noted at T12–L1 junction and L2. Extensive arachnoid adhesions involving the spinal cord circumferentially, nerve roots, and spinal cord vasculature were encountered, possibly related to prior hemorrhage and lumbar drain placement. After the dissection of adhesions there was a large, dorsal, arterialized vein clearly visualized. Attention was focused on the two areas seen on angiography as possible sites of fistulous connections: T12–L1 lateral cord and L2 ventral cord. At T12–L1, a tortuous nest of small vessels, apparently coming from the posterior spinal artery along the dorsal roots and lateral pia that connected to the dorsal vein, was noted. A temporary clip was applied across these vessels without change in neuromonitoring and these were cauterized and cut. The numerous and dense adhesions impaired attempts to rotate the spinal cord and directly visualize the ventral cord. A dilated vein traversing the spinal cord [Figure 1a] at the

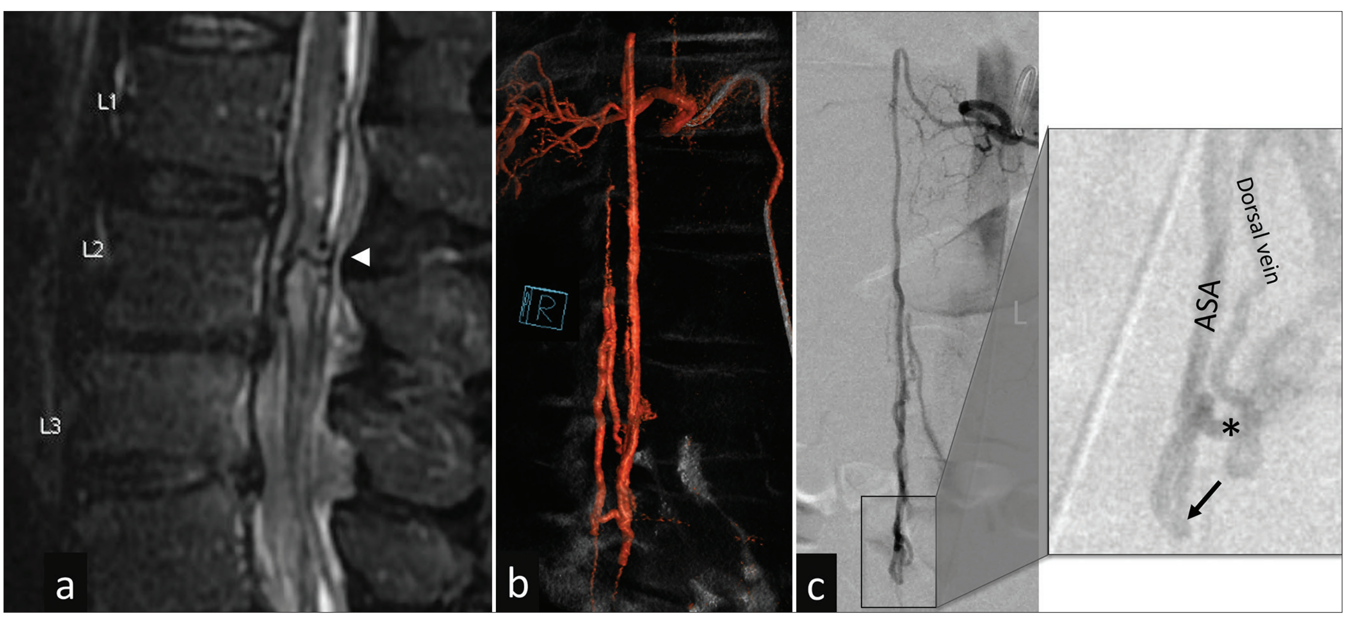

Figure 1: Preoperative images (a) T2-weighted sagittal MRI scan shows an edematous conus medullaris with a large vein crossing through the parenchyma at the level of L2 (arrowhead). (b and c) Spinal angiogram 3D reconstruction and original preoperative images. The zoom-in inset image, at a slight oblique angle, provides information on the anatomy – the dorsal vein is quickly filled with blood from main fistulous connection (black arrow) with anterior spinal artery (ASA). Smaller, superficial, feeders are not seen. The asterisk points at where the clip was applied – the intense adhesions and vessel tortuosity prevented proper clip position, which remained at a superficial (dorsal) location.

level of L2 and connecting to the dominant dorsal draining vein was carefully dissected, and no dorsal arteriovenous fistula was noted on the dorsal surface. We attempted to dissect the vessel more ventrally along the course into the spinal cord at L2, but a temporary drop in motor-evoked potentials prevented further manipulation or dissection. A clip was then applied as ventral as possible on this arterialized vein at L2 for its occlusion. Serial indocyanine green video angiography after cauterization and clip ligation showed a slowing of the vein filling after the vessel cauterization at T12–L1 and finally back to what appeared physiologic pattern after treating both areas.

Immediately following surgery patient had transient proximal left leg weakness that improved quickly to a 4/5 strength. However, on the 7th postoperative day, there was a new worsening, with bilateral, distal-predominant low extremity weakness. MRI demonstrated worsening spinal cord edema [Figure 2a]. Repeat angiogram showed a persistent SAVF with the recruitment of multiple anterior medullary veins [Figures 2b and c]. The newly recruited outflow veins were too small to be catheterized from a retrograde transvenous approach, and the previously hypertrophied vein was now occluded by a surgical clip [Figure 2c]. Therefore, the reintervention with the described novel technique was planned. Given the declining neurological examination, the urgency of intervention, and the limitations of standard surgical-only or endovascular-only treatments, no Institutional Review Board or Ethics Committee Review was pursued. The patient consented to a hybrid open-endovascular approach.

The patient underwent a hybrid open-endovascular approach involving the removal of a previously placed clip on the hypertrophied vein and puncture of the large dorsal vein with a 20-gauge Angiocath. A 1.3 French Headway Duo 167 microcatheter (Microvention Inc, Aliso Viejo, CA, USA) was advanced over a Chikai 008 microwire (Asahi Intecc USA, Tustin, CA, USA) and retrograde access was obtained through wire recanalization of the thrombosed, previously clipped vein [Figure 3a]. Direct catheterization of the fistulous point was attempted to perform coil embolization as this was felt to be safer. Due to tortuosity, the microcatheter could not be advanced further, and the decision was made to use a liquid embolic to reach the fistulous point. The thrombosed outflow vein acted as a backstop to prevent reflux. Embolization with Onyx 18 (Medtronic Neurovascular, Irvine, CA, USA) was performed, and the embolic was advanced until all outflow veins disappeared on transradial spinal angiography [Figures 3b ,c]. Onyx 18 liquid embolic is labeled for presurgical embolization of brain arteriovenous malformations and not specifically for use in SAVFs. After surgery, the patient's progressive neurological decline arrested, radiographic spinal cord edema improved [Figure 3d], and follow-up angiography confirmed obliteration of the fistula [Figures 3e and f]. Unfortunately, neurological function remained with none or minimal improvement, with significant residual deficits in lower extremity strength, sensation, and sphincter dysfunction.

DISCUSSION

In this case, open surgical access provided a route for surgical closure of dorsal and laterally located fistulous connections and provided successful transvenous endovascular access to the ventral feeder [Figure 4]. A direct percutaneous injection

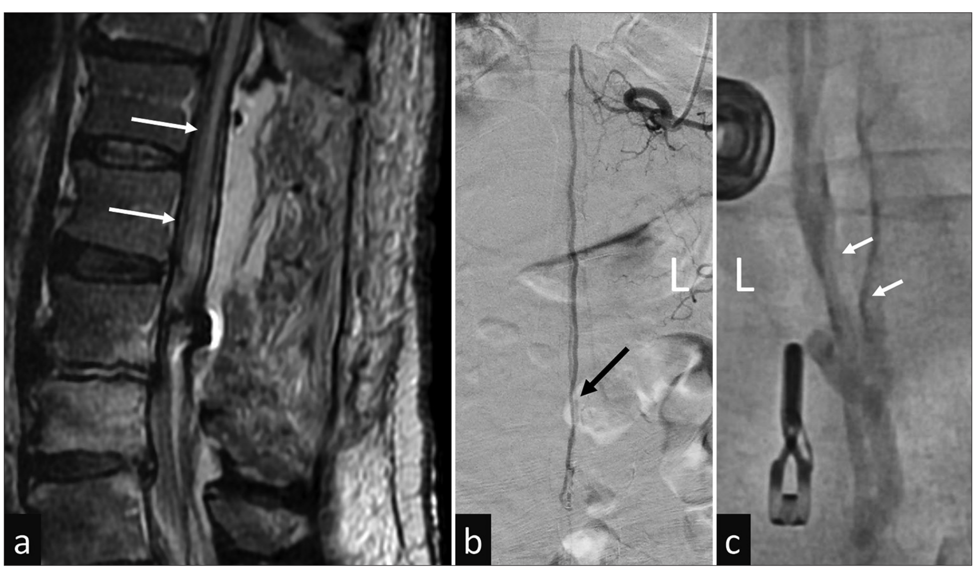

Figure 2: Post clipping images (a) T2-weighted sagittal MRI scan after initial fistula treatment demonstrates increased cord edema extension (arrows). (b) Control angiogram demonstrated recruitment of multiple anterior medullary veins after clipping (black arrow). (c) These recruited veins, easier viewed at a higher magnification, are pointed out by the white arrows.

for embolization[15] was felt not feasible given the modest size of the veins and the direct use of glue in the traversing venous system without fluoroscopy guidance can damage any functional vascular supply to the cord.[2] The initial open approach with clip ligation of the dorsal draining vein at the dorsal pial surface likely failed as it did not block off the SAVF at the ventral pial surface or point of fistulous connection and allowed redirection of flow to additional, smaller venous outflows that were not initially visualized on angiography. Our technique combining a direct venous puncture with

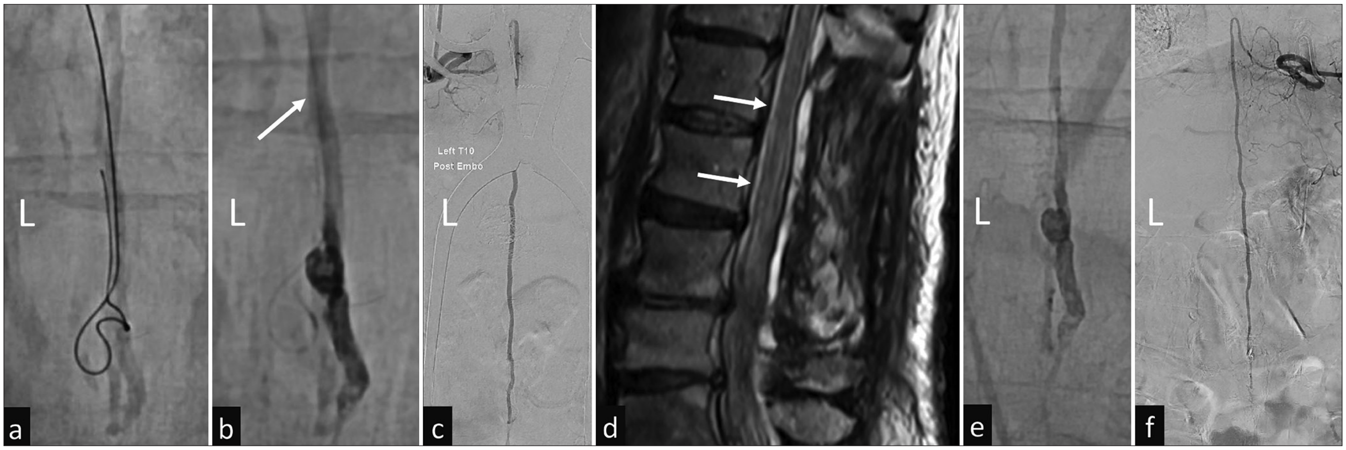

Figure 3: Figure 3: Intra and postembolization angiograms and MRI. (a) Intraprocedural angiogram shows the advancement of the catheter through the thrombosed segment after clip removal. (b) Immediate post onyx embolization demonstrating occlusion of the feeding vessels with white arrow pointing to the puncture site in the dorsal vein. (c) Intraoperative angiogram immediate post-embolization. (d) MRI after revision hybrid open-endovascular treatment demonstrates decreased spinal cord edema (arrows). (e and f) 1-month follow-up angiography with stable findings and no arteriovenous shunting.

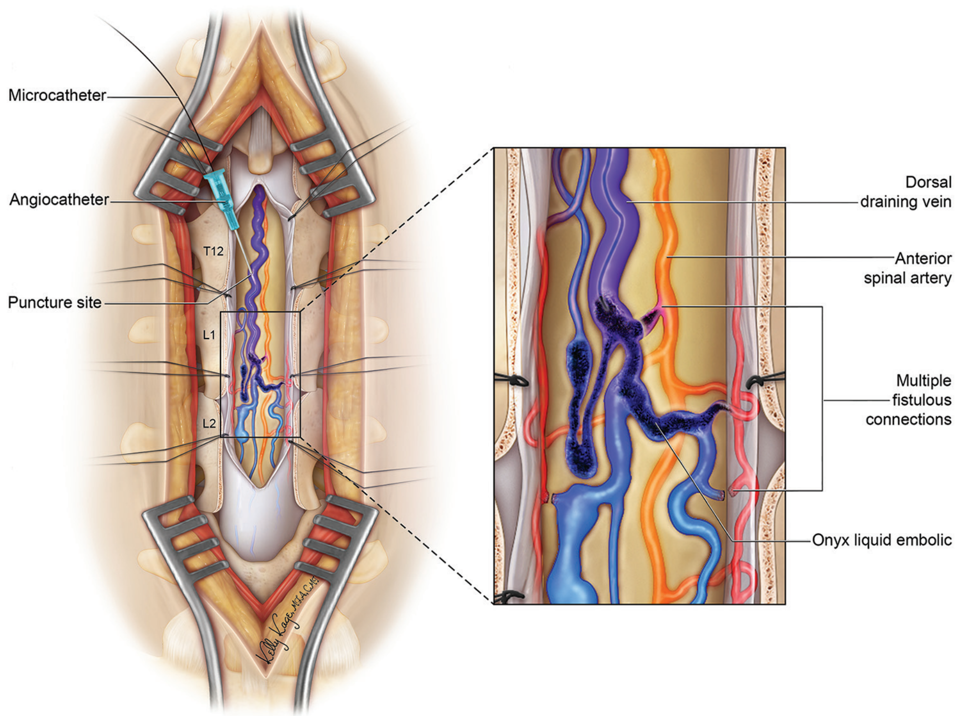

Figure 4: Artistic representation of the hybrid approach to the spinal arteriovenous fistula. In this representative case, based on the actual case here presented, the spinal cord is exposed through a standard posterior approach. Dorsal and superficial feeders can be coagulated and cut. Deep feeders, crossing through the spinal cord, can be reached by the advance of microcatheter through the engorged recipient dorsal vein and occluded with liquid embolic.

a microcatheter under fluoroscopic guidance allowed for closer and more controlled transvenous access to the fistulous pouch in the ventral aspect of the spinal cord. Trans-arterial access may have allowed for successful treatment, but this approach was felt at high risk for spinal cord ischemia and was not attempted. Although our patient did not recover his neurological function, further neurologic decline did not occur. The technical limitations for these hybrid open and endovascular approaches in the spine include the following: the access to an adequate angiography and surgical equipment in the same operating room, stabilization of the Angiocath during access and microcatheter advancement to avoid accidental loss of access or damage to the spinal cord; working in a deep surgical cavity with limitations in hand movement during endovascular treatment; soft-tissue retractors and operating room equipment that obscure the surgical and/or fluoroscopy view; and proximity of the hands of multiple surgeons holding the equipment that may obscure the fluoroscopy view and lead to increased radiation exposure of the surgeons.

CONCLUSION

This hybrid approach may be a treatment for selected cases of type IVb SAVF. The multiple feeding vessels can be addressed according to their accessibility: easily surgically accessible ones are coagulated and cut, whereas the inaccessible ones can be endovascularly accessed and embolized during the same procedure.

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Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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