A treatment-refractory spinal dural arteriovenous fistula sharing arterial origin with the Artery of Adamkiewicz: Repeated endovascular treatment after failed microsurgery

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Abstract

Background: Effective management of a spinal dural arteriovenous fistula (SDAVF) can be accomplished with either microsurgery or endovascular embolization, but there is a consensus that in patients in whom a radiculomedullary artery supplying the anterior spinal artery (ASA) originates from the same feeding artery as the SDAVF, the endovascular approach is to be avoided.

Case Description: The patient was a 46-year-old woman with progressive lower limb paraparesis, sensory deficit, and sphincter dysfunction. Magnetic resonance imaging (MRI) and spinal angiography showed an SDAVF fed by a branch from the left second lumbar segmental artery, and the artery of Adamkiewicz (AA), a major ASA supplier, originating from the same segmental artery just proximal to the SDAVF. Microsurgical disconnection of the SDAVF was attempted, but failed. Embolization with cyanoacrylates was done in two occasions, the first time through a microcatheter placed just distal to the origin of the AA and the second time through another feeder coming from the same segmental artery that could not be visualized in the previous angiographies. All procedures were neurologically uncomplicated. Magnetic resonance imaging (MRI) 1 month after the last embolization showed resolution of the spinal cord edema. MRI scan taken 68 months after embolization revealed a slightly atrophic spinal cord with visible central canal and no recurrence of medullary edema. The patient presented good, but incomplete neurological improvement.

Conclusion: Microsurgery is the first choice for an SDAVF branching off the same radiculomedullary artery supplying the ASA, but uncomplicated embolization can be feasible after failed surgery.

Key Words: Anterior spinal artery, artery of Adamkiewicz, embolization, spinal dural arteriovenous fistula, spinal surgery

INTRODUCTION

Spinal dural arteriovenous fistula (SDAVF) is a cause of progressive motor and sensory myelopathy. Therapy is disconnection of the arteriovenous fistula, which is located outside the spinal cord, by occluding the target draining vein, and can be accomplished effectively with microsurgery, endovascular embolization, or both. While
the selection criteria for each management modality may vary in different centers, it is uncontroversial that in patients in whom a radiculomedullary artery supplying the anterior spinal artery (ASA) originates from the same feeding artery as the SDAVF, embolization is to be avoided,[5] making microsurgery as the treatment of choice for these patients. This was the case with the present patient, but the arteriovenous shunt could not be removed on surgery, and subsequent endovascular embolization was done without any neurological complication.

**CASE REPORT**

A 46-year-old woman with walking difficulties, motor deficits, dysesthesia and sensory loss in the lower limbs for at least 6 years, presented with rapid neurological deterioration since 4 months, developing also sphincter dysfunction (urinary incontinence and fecal retention). She was able to walk with support.

**Imaging studies**

Magnetic resonance imaging (MRI) of the spine showed marked edema and patchy contrast agent enhancement of the spinal cord from the T7 level to the conus medullaris, and enlarged perimedullary veins predominantly on the left side [Figure 1]. Spinal angiography revealed an SDAVF in the left L2-L3 intervertebral foramen, fed by a branch from the second lumbar segmental artery (L2) on the left [Figure 2]. From the very same segmental artery, the artery of Adamkiewicz (AA), a main supplier of the ASA system, branched off about 0.5 cm proximal to the fistula. Due to this anatomical particularity, endovascular embolization of the fistula was considered contraindicated and microsurgical treatment was opted for.

**Operative procedure**

Laminectomy at the L2 level was done. Upon opening the dura mater, the veins along the nerve roots were found to be enlarged and tortuous. Two arterialized veins running along the left L2 nerve root were coagulated and cut. The intradural veins then became more bluish in color and less congested. Thus, it was deemed that closure of the SDAVF had been achieved.

**Postoperative follow-up**

The patient experienced some immediate neurological improvement, with reduced dysesthesia in the lower limbs bilaterally. She was sent for rehabilitation at the referring hospital. Three months later, follow-up MRI showed persistent medullary edema with tortuous perimedullary vessels, and the only notable difference from the pre-operative images was the evidence of laminectomy on the L2 level. The patient was re-admitted for spinal catheter angiography. On clinical examination at admission, her motor and sensory deficits were slightly improved and her sphincter dysfunction was unchanged, in comparison to before surgery.

**Spinal angiography and endovascular procedure**

On spinal angiography, the same SDAVF as before was visualized, still intact, and the AA branched off the same artery about 0.5 cm proximal to the fistula site [Figure 3a]. It was decided to attempt embolization past the branching point of the AA. The segmental artery supporting the fistula was catheterized using a 4 F catheter. A microcatheter (UltraFlow 1.5 F, used with guidewire Mirage, both from Micro Therapeutics, Inc., Irvine, CA, USA) was navigated into the fistula just distally to the origin of the AA [Figure 3a]. Embolization was done using 0.1 ml glue (50% N-butyl-2-cyanoacrylate and methacryloxyxsulfolane in Lipiodol: Glubran-2®).

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**Figure 1:** T2-weighted MRI of the spine: (a) midline sagittal image showing edema of the conus medullaris; (b) parasagittal image on the left showing engorged perimedullary veins (arrow). This finding is typical for SDAVF

**Figure 2:** Digital subtraction angiography (DSA) of the left L2 artery in anterior–posterior projection showing a dural arteriovenous fistula (lower arrow) at the left foramen L2-L3 and the AA (upper arrow: note this artery’s vertical course and typical hairpin curve into the descending branch)
GEM Srl, Viareggio, Italy, and Lipiodol® Ultra-Fluide, Laboratoire Gerbet, Aulnay-Sous-Bois, France), and the distal feeder and the fistula were filled over a length of 1 cm. The microcatheter was removed and final angiography through the guide catheter showed adequate flow to the ASA and no remaining SDAVF [Figure 3b]. Postoperatively, her neurological status remained unchanged. The patient was transferred to her local hospital for rehabilitation.

Post-embolization follow-up
The patient did not improve clinically, and dysesthesia, walking difficulties, urinary incontinence and fecal retention remained as before embolization. Follow-up MRI done 2, 6, and 11 months later showed markedly less-intensive engorgement of the spinal cord, but persistent signal changes compared to previous examinations. Since the latest MRI also showed clearly dilated perimedullary veins on the left side [Figure 4], we decided to perform a new spinal angiography.

Second endovascular procedure
Catheterization of the left L2 segmental artery was achieved using a 5 F catheter, and angiography revealed the artery of the lumbar enlargement and a persistent SDAVF arising from the same segmental artery [Figure 5a]. A microcatheter was navigated into the lumbar artery and superselective angiography done with the microcatheter just proximal to the AA permitted to discern in detail the origins of two arterial branches – one branch leading to the AA and the previously embolized feeder (which remained closed) and another more lateral and caudal branch that fed the fistula [Figure 5b]. The SDAVF was catheterized through the newly identified feeder, and embolized using 0.3 ml glue (67% Glubran-2 in Lipodol) [Figure 5c]. Final angiography of the left L2 segmental artery showed preserved flow through the artery of the lumbar enlargement without any remaining arteriovenous shunt [Figure 5d]. Angiography of the adjacent segmental arteries showed no flow to the SDAVF through these arteries.

Post-embolization follow-up
The patient could be discharged to her local hospital for rehabilitation the day after the procedure. The patient had extensive rehabilitation, and regained the ability to walk without support and managed to climb stairs and drive a car. On follow-up 6 years after the last intervention, her paraparesis improved to the point that she is able to walk 70 m without support, but uses a walker for longer distances. She has reduced temperature sensation from her knees down bilaterally, and still experiences paresthesias in the soles of her feet. She uses incontinence protection daytime and experiences urgency incontinence both daytime and nighttime.

Imaging follow-up
MRI scan taken 1 month after embolization showed complete regression of the medullary edema and the one taken 68 months after embolization revealed a somewhat atrophic spinal cord with visible central canal and no recurrence of medullary edema [Figure 6].

DISCUSSION
SDAVF, or intradural dorsal arteriovenous fistula,[7] is an acquired pathology and the most common type of abnormal spinal arteriovenous shunt.[2,5] It is usually a low-flow shunt draining in a radicular vein inside a dural nerve root sleeve. The feeder artery, usually a radicular artery supplying only the dura mater and nerve root, is, as a rule, small and tortuous. Arterialization and increased pressure in the coronal venous plexus and impaired venous drainage on the spinal cord over several segments lead to progressive motor and sensory...
myelopathy that can evolve into necrotizing myelopathy over time. SDAVF type A has a single feeding artery and type B has two or more feeding arteries converging and forming a single intradural fistula. SDAVF is located most often in the midthoracic and thoracolumbar spine, but it can occur at any level from the sacrum to the foramen magnum. There is a male predominance, and the patients are typically elderly. Symptom progression is usually gradual, but in some patients, it is associated with episodes of acute-onset neurological events. Hemorrhage from an SDAVF is very rare and associated with acute clinical deterioration. Diagnosis with magnetic resonance imaging (MRI) is straightforward: The spinal cord is enlarged, shows central edema, may present patchy contrast agent enhancement, and there are multiple serpentine veins on its surface. Selective spinal angiography is essential to confirm diagnosis and direct treatment planning. The progressive myelopathy can only be halted by occluding the arteriovenous shunt, which can result in neurological improvement depending on the degree of previous disability (as a rule, motor function improves first followed by sensibility, and sexual dysfunction and loss of sphincter control are less likely to improve).

In the management strategy for SDAVF, identifying the relation of the arteriovenous shunt and radicular arteries supplying the ASA is crucial. The risk of closure of the ASA has led to a general agreement that fistulas branching off the same vessel as an ASA supplier should be treated primarily by open surgery rather than through embolization, due to the devastating effect an accidental closure of the ASA would have.\[^{1-4}\] The spinal cord is supplied by three longitudinal arterial axes:\[^{2,8}\] the single ASA (extending in the midline from the foramen magnum to the filum terminale, it originates at the vertebral arteries and is reinforced by a varying number of anterior radicular arteries of which the main and sometimes only one is called the AA, or artery of the lumbar enlargement, and supplies the anterior two thirds of the spinal cord) and two posterior spinal arteries (arising on each side from the vertebral artery or from the posterior inferior cerebellar artery, they are reinforced by a varying number of posterior radicular arteries). While accidental occlusion of a posterior radicular artery during therapy usually has no major clinical consequences, occlusion of an anterior radiculomedullary artery may lead to severe spinal cord lesion. Given SDAVF predominance in midthoracic and thoracolumbar sites, identifying any relation of the arteriovenous shunt to the AA is crucial. This major ASA-supplying artery has its origin at T9-T12 levels in 75% of the cases, and is more common on the left side.

To our knowledge, there is one earlier rapport of attempted transarterial embolization of an SDAVF arising from the same segmental artery as the AA.\[^{1}\] As in our case, the segmental artery involved was the left L2 artery, which could be catheterized just beyond the ASA feeder, but the procedure was interrupted as the embolic agent (N-butyl cyanoacrylate and Pantopaque in the ratio 34:66) hardened within the microcatheter. The catheter system was withdrawn, and the patient underwent laminectomy and successful SDAVF closure.

Concerning treatment, in most cases of SDAVF, open surgery and endovascular techniques are fully interchangeable; thus, the selection criteria for microsurgery or embolization may vary in different centers. In our institute, embolization is the first therapeutic alternative for SDAVF, and is done in the same occasion as the diagnostic spinal angiography, whenever it is considered feasible and anastomoses between the feeder artery and the ASA system are excluded. In the present case, the choice of microsurgery...
was uncontroversial, and the fact that the fistula was not occluded at the primary surgical intervention was rather unfortunate, since the surgeon was highly experienced and the correct level was operated on (postoperative MRI showed the laminectomy was performed as planned on the L2-L3 level), but still the fistula was not occluded. The coagulated and severed vein was probably only part of the enlarged venous network caused by the fistula and not the single draining vein. At reoperation, adherences were to be expected around the SDAVF and around the AA as well; thus, the risk of repeated surgery seemed to be considerably higher than that of the first operation. On the other hand, the fistula as well as the AA were well-defined on angiography and precise superselective catheterization was feasible; thus, we considered it appropriate to embolize the fistula with glue. It was necessary to repeat even the endovascular procedure, and in this respect, two comments are pertinent. First, the SDAVF was of type B with two feeders coming from the same segmental L2 artery, but one of the feeders was angiographically hidden at the time of the first embolization. Second, it is arguable that a more aggressive glue injection in the SDAVF would have effectively occluded a longer length of the draining vein (as was done in the second embolization), but even retrospectively, the interventionist considers the risk of reflux of glue to the ASA should not be underrated.

CONCLUSION

It is not our intention to recommend that an SDAVF branching off the same segmental artery as a major ASA supplying artery should be treated primarily by embolization, but merely to describe that with modern endovascular technique and great care taken not to allow reflux of embolic material, such treatment is feasible. It was fortunate that the anatomy of the fistula allowed for safe catheterization and embolization, but not all patients can be expected to display such favorable conditions. Surgical treatment, as it was in this case, remains the primary choice of treatment for similar lesions at our center.

REFERENCES
