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## Management of spinal dural arteriovenous fistula

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**Abstract:** Spinal dural arteriovenous fistulas are rare vascular lesions whose management is still at high interest between specialists. If microsurgical treatment is still considered as treatment of choice for SDAVFs, endovascular treatment is increasingly growing in interest with the development of endovascular techniques and new embolization materials. In this article we made a short discussion about the spinal dural arteriovenous fistulae on aspects related to anatomy, pathophysiology, diagnosis and treatment, with some general conclusions.

**Key words:** spinal dural arteriovenous fistulas, endovascular treatment, microsurgical treatment

### Introduction

Vascular malformation of the spinal cord represents a rare clinical condition characterized by a difficult diagnosis and complex management. Spinal dural arteriovenous fistulas (SDAVF) are the most common injuries in these pathological entities with important clinical implications. These direct communications between radicular artery and medullary vein usually result in myelopathy due to venous hypertension effect. With the advances in neuroimaging, microneurosurgery and neuroendovascular techniques the complete treatment of these pathological situations is very feasible with the possibility of complete remission of clinical symptomatology. Endovascular embolization was reported as

an effective therapy in the treatment of SDAVFs that can be used as singular and definitive intervention in some particular cases. We present a particular case with SDAVF treated by endovascular embolization and discuss the treatment possibilities to more fully understand the optimal management of these lesions.

### Vascular Anatomy

Spinal cord vascularisation is provided by the anterior spinal artery (ASA) and the paired posterior spinal arteries (PSA). The ASA consists of the junction of two branches originating from the two vertebral arteries proximal to the vertebrobasilar junction. On its path, it receives contributions from branches of vertebral and ascending cervical

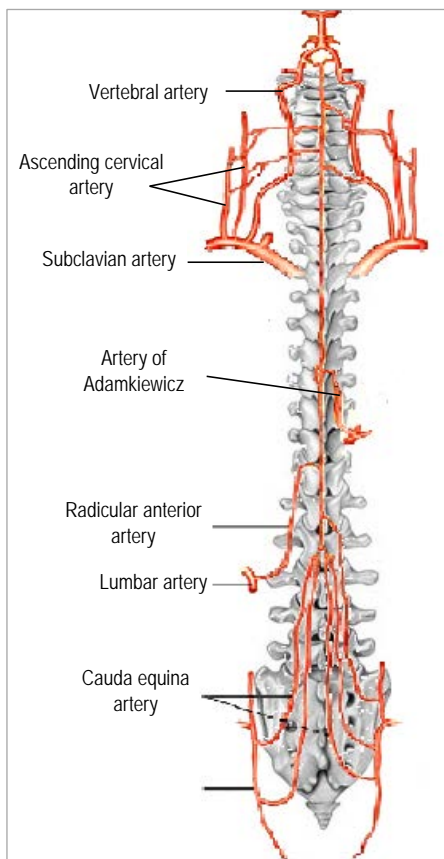
arteries in the cervical region as well as from intercostal and lumbar arteries at the corresponding levels. The radiculomeningeal arteries are branches of the segmental arteries founded at almost every spinal level supplying the dura in the spinal canal. Unlike these, radiculomedullary arteries, which exist only at some levels, are implicated in spinal cord vascular perfusion. The artery of Adamkiewicz (great anterior radiculomedullary artery) is the dominant thoracolumbar segmental artery with variable origin from T8 to L1 vertebral segments that connects to ASA and supplies the spinal cord. The posterior spinal arteries arise from either the posterior inferior cerebellar or vertebral arteries (V3 or V4 segments) and as they descend on either side of the dorsolateral cord surface they are reinforced by segmental/radicular branches. It anastomoses with its fellow and with the anterior spinal artery [1,6].

### **Epidemiology and Pathophysiology**

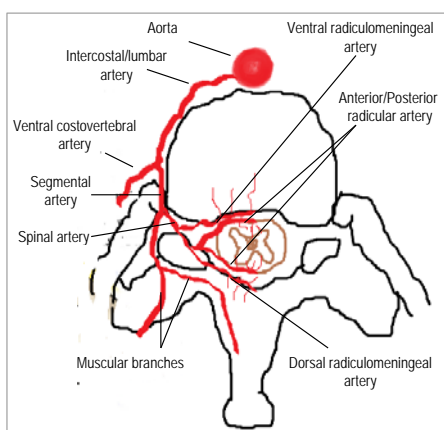
SDAVF represents 70% of spinal arteriovenous shunts that commonly occur in the thoracic and lumbar spines of middle-aged men[4,6]. The majority of SDAVFs occur spontaneously, but a post-traumatic etiology cannot be excluded in a significant proportion of them. Typically, this disease affects male patients (in 80% of cases) in their 50s and 60s,<sup>3</sup> The pathophysiology of SDAVF consist in spinal cord venous hypertension due to one or a few small low-flow arteriovenous shunts between a radiculomeningeal artery and a radiculomedullary vein, typically located in

the intervertebral foramen within the dura[4,6,7].

The retrograde venous drainage circuit in SDAVF is represented by a radiculomedullary vein (most frequently dorsal to the cord) into the perimedullary venous system and finally the medullary veins. The venous drainage of the SDAVF is slow and expansive, and may reach the cervical spinal canal or cauda equine by ascending or descending blood reflux. Because the radiculomedullary veins are not anatomically numerous the presence of SDAVF is often associated with their and epidural veins congestion and thrombosis. That explains why the low-flow arteriovenous shunt of SDAVF induces a rises of venous pressure (74% of the mean arterial pressure) which leads to decreased arteriovenous gradient, segmental spinal cord edema that may progress to congestive ischemia and necrotizing myelopathy. The caudocranial progression is favored by a valveless venous system of the cord resulting in 'arterialization' of these veins with thickened and tortuous walls. The pressure in the draining vein also varies with arterial pressure and may lead to an accutization of simptoms. Because the SDAVFs is a slow-flow fistulae, hemorrhage is a rare clinical manifestation. Subarachnoid hemorrhage is rarely encountered especially in high cervical localization. [6,7]



**Figure 1**



**Figure 2**

## Classification

Many classification systems were reported and changed over time in the literature. Between 1971 and 2011 seven major classification systems have been enunciated based on the evolution of diagnostic methods and treatments for spinal AV shunts. The most used described in 2002 by Spetzler and colleagues and divided the vascular spinal lesions in SAVFs and SAVMs. SAVFs are further subdivided based on their extradural versus intradural location. The intradural SAVFs were divided in ventrally or dorsally due to their relation to the spinal cord. In turn, intradural ventral SAVFs are further divided into types A, B and C depending on the number and size of feeding branches [1,6].

Extradural SAVFs represents direct connection between a branch of a radiculomenigeal artery and the epidural venous plexus (Figure 4). These are rare entities characterized by enlargement of epidural veins with medullary venous congestion that may cause compression of the spinal cord or nerve roots. More recently Rangel-Castilla et al. divided these lesions in type A (SAVFs drain into both the epidural venous plexus and perimedullary venous plexus) and type B (SAVFs drain only into Batson's plexus). Type B1 lesions compress the thecal sac due to an enlarged epidural venous plexus and type B2 lesions lack such compression [1,6,7].

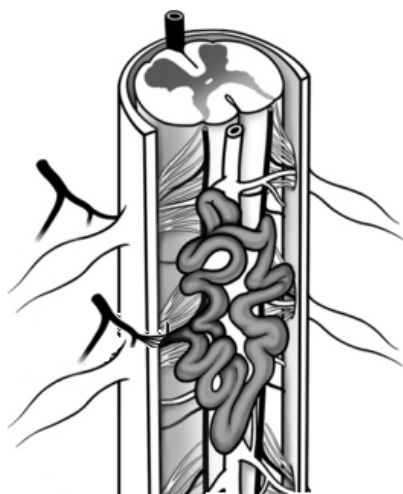


Figure 3

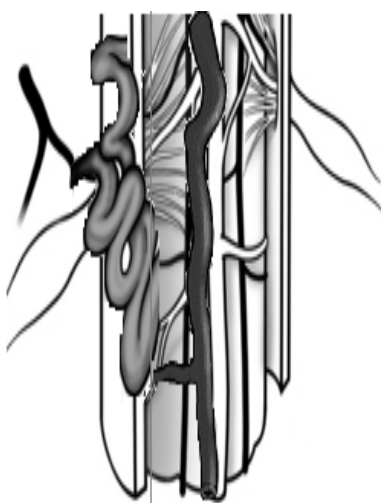


Figure 4

Intradural dorsal SAVFs are the most common type of spinal vascular malformation consisting in a direct connection between a dorsal radiculomedullary artery and a medullary vein at the dural nerve root sleeve (Figure 3). Progression of venous hypertension to the coronal venous plexus leads to venous congestion and progressive myelopathy.

Intradural ventral SAVFs are typically high-flow direct fistulas between the ASA and coronal venous plexus. The lesions develop in the ventral subarachnoid space and can be further categorized into three subtypes according to their size. Type A fistulas are single-feeder lesions with slow blood flow and mild venous hypertension. Type B fistulas are progressively high-flow lesions with multiple minor feeders. Type C fistulas are usually large fistula with a markedly enlarged venous drainage [1].

### Clinical symptoms

Most clinical reports showed a delay between the onset of clinical symptoms and diagnosis of these vascular lesions (between 12 and 44 months) [6]. This is largely due to nonspecific clinical presentation. The presenting symptoms usually include a combination of unilateral or bilateral lower extremity motor weakness that is worsening by intense movements. Gait disturbance, sensory symptoms (pain, paresthesias, diffuse or irregular sensory loss, hyperesthesia) and sphincter/bladder disturbances are also seen and commonly lead the clinicians to consider or exclude many other disorders before considering SDAVFs. Often misleading, mono or polyradiculopathy and low back pain are encountered and contribute to the difficulty of true diagnosis. Bowel and bladder incontinence, sexual dysfunction and urinary retention are seen late in the course the disease process. The symptoms are typically progressive and the natural evolution of untreated patients is to severe aggravation over a period of 6 months to 2 years. Spontaneous

recovery has not been reported so far as sudden worsening has been more and more common [1,7].

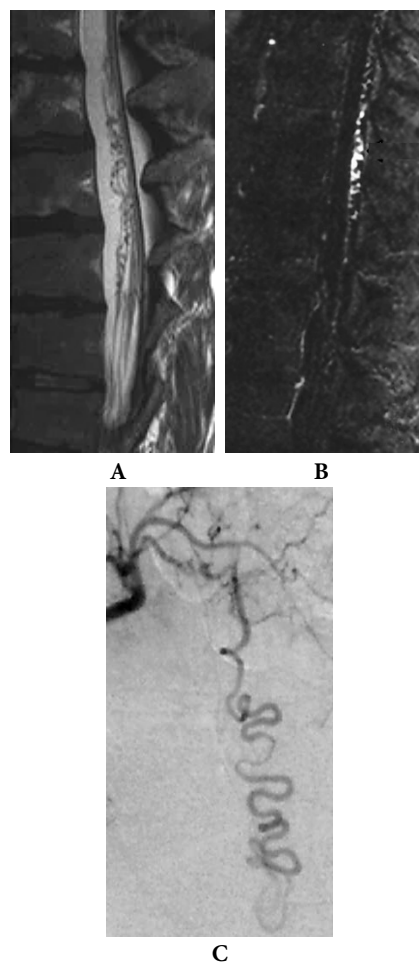
Misdiagnosis usually includes degenerative spine diseases, spinal cord tumours, neuromuscular diseases, peripheral vasculopathy or neuropathy.

### Imaging Diagnosis

It is all accepted that the MRI is the primary investigation for the evaluation of myelopathy and the first line in diagnosis of SDAVFs. The presence of dilated perimedullary serpentine vessels on T2 signal was found to be the most sensitive MRI findings in SDAVFs. The abnormal shunts are much better outlined after administration of intravenous gadolinium-based contrast agents, increasing the sensitivity and specificity of MRI exam for SDAVFs diagnosis (Figure 5A). The MRA sequences permits a better imaging characterization of the abnormal enlarged perimedullary vessels concerning their relative size, number and tuotuasity. MRA is a useful tool for planning the surgical treatment and following monitoring (Figure 5B). Other imaging signs like spinal cord edema, enlargement, gliotic or atrophy were reported depending on the stage of venous hypertension [6].

Conventional catheter angiography is the gold standard investigation for the diagnosis and classification of the SDAVFs. The selective catheterization and evaluation of each individual segmental arteries at thoracic and lombar level have to be performed for SDAVF identification. Once an identified fistula, a prolonged angiographic imaging

acquisition has to be performed for a complete characterization of the venous drainage (Figure 5C). In patients with sever venous hypertension and myelopathy at thoracolumbar area the venous drainage is delayed or even absent. For suspected cervical or lombosacral SDAVFs the angiographic investigation of vertebral and ascending cervical arteries or the internal iliac and iliolumbar arteries should be performed.



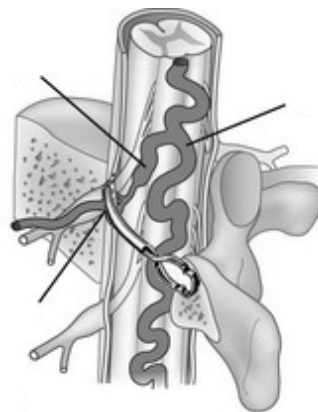
**Figure 5** - SDAVF aspects on A - MRI - T2; B - MRA and C - Catheter angiography

## Treatment

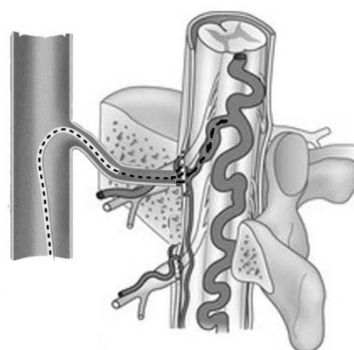
Treatment of SDAVF must be performed as soon as possible and it could be microsurgical ligation, endovascular obliteration, or both. Although the microsurgical treatment is considered the gold standard technique, the endovascular treatment could be a feasible and safe option.

The microsurgical approach is performed by a posterior approach with a midline laminectomy one level above and below the fistula origin. The dura is opened in the midline and the radiculo-meningeal artery shunt must be identified extradural. In the case of a fistula with multiple small arterial pedicles, these should be carefully identified by dissection along the dural root sleeve (Figure 6). The draining arterialized vein must be exposed and clearly identified against dilated perimedullary veins. The microsurgical technique consists in cauterization and microscissor interruption of the fistula. Postoperative angiography is indicated to confirm complete surgical obliteration. Most of the studies have reported an improved disability scores and lower recurrence rates after microsurgery treatment compared with endovascular obliteration. Surgical management of SDAVF was also necessary when an incomplete endovascular obliteration or recanalization were the final results [2,3].

Once the improvements in endovascular technique and embolic materials, the endovascular treatment of SDAVFs has more largely used.



**Figure 6** - Microsurgical clipping and disconnection of artery feeder



**Figure 7** - Endovascular glue injection in artery feeder and proximal radiculomedullary draining vein

The success of endovascular treatment was considered when a complete occlusion of the proximal radiculomedullary draining vein and the site of the fistula itself were obtained. The procedure consists in right transfemoral access by 6F sheath placement. After the identification of the arterial supply of SDAVF a guiding catheter is placed at the ostium of the corresponding segmental artery to offer more support for navigation into the often tortuous feeders. Then a microcatheter is advanced under road-mapping over a

microwire in order to reach the closest point to the fistula. If the embolization is performed too proximally in the radicular feeding artery, collateral feeders could develop distally and re-permeabilized the fistula. A microcatheter angiography is recommended before starting the embolic agent injection in order to ensure if the anterior spinal, posterior spinal or a radiculomedullary artery are not directly connected to fistula. Endovascular occlusion is performed by slowly Glubran 2 or Onyx injection into proximal draining vein while occluding the fistula site and feeding arterial vessels (Figure 7). Finally, control angiography is performed by selective catheterization of the segmental arteries arising at least two levels above and below the SDAVF site. If there is no complete obliteration of the fistula usually the patient is addressed for a microsurgical approach as soon as possible. Also, if there are doubts concerning the complete occlusion of the proximal radiculomedullary draining vein, a control angiographies are performed at one, three and six months later[2,3,6].

### Illustrative Case

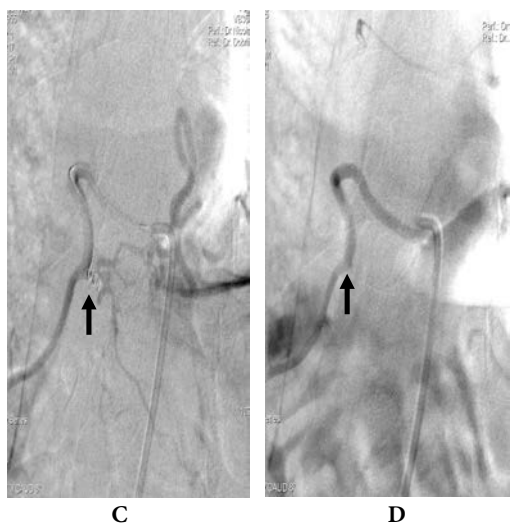
#### Case 1

This 62-year-old male presented with progressive gait instability, numbness and dysesthesias in the bilateral lower extremities, as well as increased lower extremity fatigue and urinary incontinence. Magnetic resonance imaging and MR angiography of the spine demonstrated diffuse cord edema in the dorsal spine, with multiple abnormal blood vessels surrounding the spinal cord. These findings prompted a referral of the patient to our

neurosurgical department for further evaluation and treatment of a suspected SDAVF. Spinal angiography was performed by selective injection of the right T-10 intercostal artery. The selective angio showed filling of a right radiculomeningeal branch from the region of the nerve root sleeve that filled a fistula (Figure 8A, 8B). No other contributions to the spinal SDAVFs were identified. The ASA was found to arise from the left T-11 intercostal artery with no implication on fistula. The right radiculomeningeal branch is then microcatheterized and after detachment of a GDC-10 coil at its level the feeding branches were embolized using Glubran 2. (Figure 8C, 8D) Control angiography demonstrated complete obliteration of the SDAVF without compromise to the right T-10 intercostal artery. One week following embolization procedure, his gait began to markedly improve and he was discharged to recovery clinic.





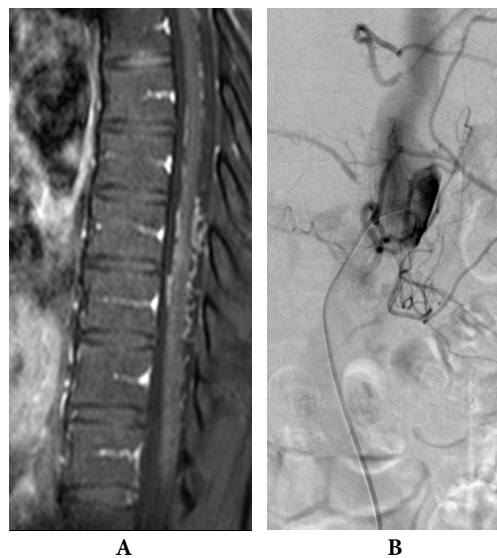


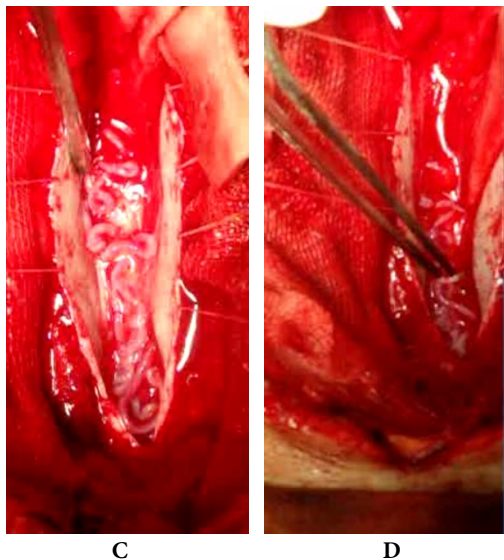
**Figure 8** - T-10 SDAVF A - MRI - T2; B - Catheter angiography; C - GDC-10 detachment; D - SDAVF occlusion

### Case 2

A 61-year-old male was addressed to a neurologic department for progressive gait instability. Magnetic resonance imaging of the spine demonstrated multiple abnormal blood vessels surrounding the spinal cord (Figure 9A). Based on the suspicion of a spine AVM the patient was addressed to our neurosurgical department. At admission the neurological examination revealed bilateral lower-extremity motor weakness and sensory deficits, with the left side more severely affected. There was no motor or sensory deficit in the upper extremities. Selective spinal angiography at left T-7 intercostal artery showed SDAVF supplied by its radiculomeningeal branch and draining into a tortuous proximal radiculomedullary vein (Figure 9B). A posterior approach with a midline T7-T8 laminectomy was performed.

The dura was opened in the midline, the radiculomenigeal artery shunt was identified and a clip is applied on it. If the collapse of the drainage vein is observed with the change of blood flow arterial arterial to venous, the shunt is cauterized and disconnected (Figure 9c, 9D). Postintervention angiography was performed for fistula interruption documentation. The patient was discharged to a local recovery centre. He continued to improve and was almost back to his baseline neurological and ambulatory status at the 6-month follow-up.





**Figure 9** - T-7 SDAVF A - MRI - T1/TSE/ENHANCMENT; B - Catheter angiography; C - Shunt identification; D - Shunt cauterization and micro scissor disconnection

## Discussion

SDAVFs were defined as abnormal direct connection between a radicular extradural artery and an intradural vein. Most of studies show a predominance of the lesion in the thoracic spine and to male gender. The vast majority of patients presents with different degrees of neurological impairments usually correlated with the level of venous hypertension and its time occurrence.

Many authors are still considered the surgical obliteration of SDAVF to be the gold standard for management of these lesions. However, improvements in endovascular technique and development of new embolic materials have made a greater number of patients with such vascular lesions to be treated for this type of treatment. The

literature presents rates of successful endovascular therapy that vary between 25% and 90%[4,7]. The advantages of this treatment have been associated with shorter time hospitalization, minimal procedural morbidity and earlier initiation of rehabilitation programs. Contraindication for endovascular occlusion o SDAVF are represented by the spinal cord supply from the same arterial trunk as the feeding artery of the fistula, difficulties of a distal catheterization of the feeder artery due to its anatomical particularities, or recanalization of fistula after a previous embolization session. All experts have agreed that the success of endovascular treatment is closely related to the complete occlusion of both the arterial feeder and the proximal radiculomedulary vein.

Recent comparative studies between microsurgical and endovascular treatment of SDAVFs on larger series of patients have shown that there are no statistically significant differences in postinterventional neurological recovery. Early diagnosis and successful treatment of the fistula were demonstrated to be strong correlated with improvement in clinical symptoms. It was also found that improvement in motor function after treatment is more likely to occur than improvement in urinary dysfunction. The patients must be postinterventionally monitored by clinical examination and at least MRI imaging. In endovascular treated SDAVFs a catheter angiography control is recommended.

## Conclusions

Endovascular treatment of SDAVFs represents a good and effective option for management of these vascular lesions. However, some limitations on the possibility of applying this type of treatment have been described. For most cases, surgical treatment is still considered the first intention treatment.

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