Spinal Dural Arteriovenous Fistula with Bilateral Lateral Sacral Arterial Supply and Embolization

David J. Lerner, M.D.[1], Michel Samson, MS3[2], William Holloway, M.D.[3], Naveed Akhtar, M.D.[3]
[1]University of Missouri-Kansas City School of Medicine, Kansas City, MO
[2]Saba University School of Medicine Saba, Dutch Caribbean
[3]St. Luke's Hospital, Kansas City, MO

Introduction

Spinal dural arteriovenous fistulas (SDAVFs), although rare (5-10/1,000,000), are the most common type of spinal vascular malformation.[1] They have a significant lag time in initial onset of symptoms to diagnosis with significant morbidity such as paraplegia and loss of bladder or bowel control. They are almost always treatable and preventable of progression upon diagnosis. SDAVFs usually arise from the thoracolumbar vertebral arteries, and rarely arise from the lateral sacral artery. We present a case of a spinal dural arteriovenous fistula arising from bilateral lateral sacral arteries with curative treatment from neuro-interventional radiologic embolization.

Case Report

A 75-year-old female presented to the emergency department for a chief complaint of progressive bilateral lower extremity myelopathy of four years with recent significant worsening. Over the course of four years, she reported progressive back pain, paresthesias, and bilateral lower extremity weakness necessitating the use of a cane progressing to needing a four-wheeled walker. She had become even weaker recently almost to the point of being unable to move her bilateral lower extremities as well as loss of bladder and bowel function.

Magnetic resonance imaging revealed numerous tortuous small vessel flow voids over the surface of the spinal cord as well as cord edema in the lumbar and thoracic spine (see Figure 1). Given the patient's history, these findings were representative of spinal dural arteriovenous fistula. The patient was transferred to the institution's main hospital for diagnostic angiography and possible intervention.

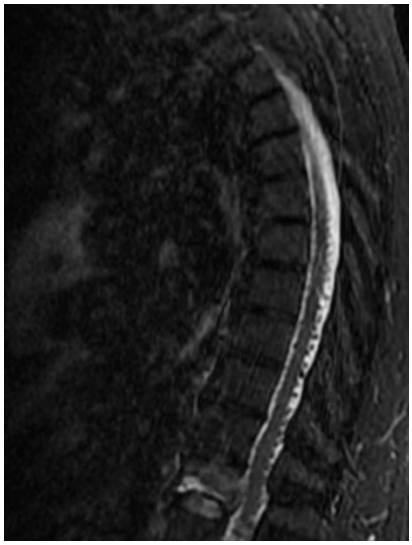

Figure 1. Saggital T2 Short TI Inversion Recovery (STIR) sequence displays cord edema together with perimedullary dilated vessels without intramedullary nidus characteristic of a SDAVF. Incidental note is made of vertebroplasty.

Catheter-directed angiography was performed in the neuro-interventional radiology suite with right femoral access (see Figure 2). Sub-selection and manual contrast injection of bilateral thoracolumbar vertebral arteries was performed. The artery of Adamkowitz was identified. The catheter was retracted to the level of the common right femoral artery and power contrast injection of 25 ml/sec for a maximum of 50 ml/sec was performed with contrast reflux into the aorta. A left lateral sacral artery supplied SDAVF ascending superiorly along the midline of the spinal canal was revealed.

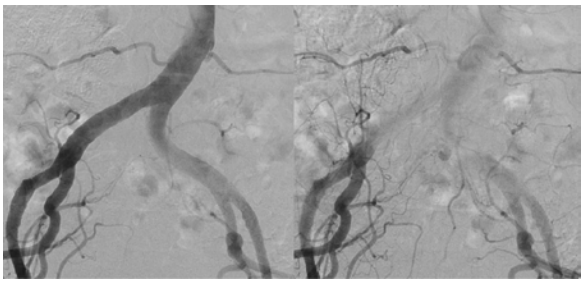

Figure 2. Left: Digital subtraction angiography (DSA) of pelvic angiogram shows an abnormal vessel arising from the left lateral sacral artery. Right: Delay DSA image of same run displays abnormal vessel not washing out with further superior extension.

The patient received catheter-directed embolization under anesthesia on the following morning. Microcatheter sub-selection of the left lateral sacral artery was obtained with re-demonstration of the SDAVF (see Figure 3). N-Butyl Cyanoacrylate Glue (NBCA) was mixed with opacification material and injected into the distal-most aspect of the left lateral sacral artery (see Figure 4). The catheter was retracted to the right common femoral artery and contrast injection was refluxed into the aorta. A subtle opacification of the SDAVF from a right lateral sacral artery feeding vessel was revealed (see Figure 5). Microcatheter angiographic sub-selection and glue injection of this artery was performed. A final run displayed no opacification of the SDAVF. The patient tolerated the procedure well. Over the course of the next few days, she regained her ability to ambulate and improved her bladder and bowel control.

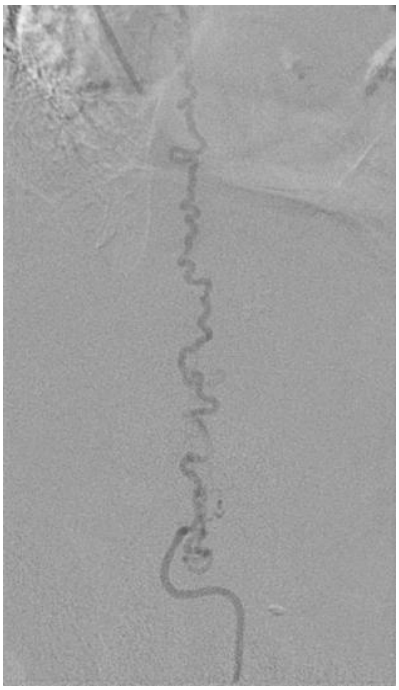

Figure 3. DSA image after sub-selection of left lateral sacral arterial malformation and subsequent contrast injection displays tortuous vascular anatomy over the location of the lumbar spine consistent with spinal dural arteriovenous malformation.

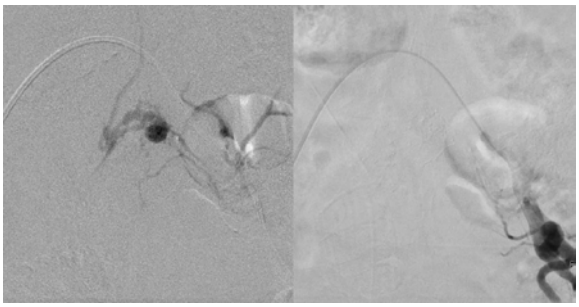

Figure 4. Left: DSA image of pelvis shows injection of opacification material mixed glue into the left lateral sacral feeding artery. Right: DSA run of same artery after glue release shows non-visualization of SDAVF consistent with technical success.

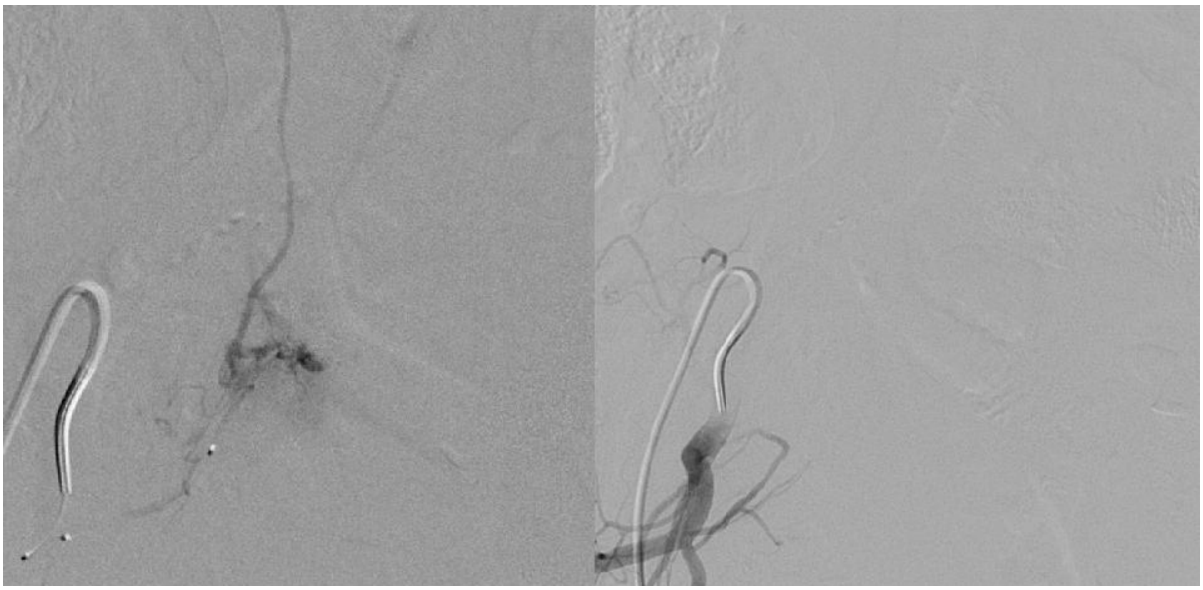

Figure 5. Left: DSA image shows contrast injection of right lateral sacral artery which displays additional feeding vessel to the SDAVF. Right: DSA image shows right lateral sacral artery contrast injection status post glue embolization which displays non-visualization of SDAVF consistent with technical success.

Discussion

Spinal dural arteriovenous fistulas are the most common vascular malformation of the spinal cord.[1-4] They account for approximately 80% of all spinal AV malformations. The underlying pathophysiology of SDAVFs is unknown, however, the pathology is usually the result of a radicular artery and vein that are abnormally connected. This abnormality usually occurs at the intervertebral foramen near the nerve root typically between L6 and T2 accounting for roughly 80% of all SDAVFs. Sacral SDAVFs accounted for only 4% of cases (5-10/25,000,000).[1-4] Sacral SDAVFs that have feeding arteries from bilateral lateral sacral arteries are rarer with less than 100 reported cases in PubMed.[4]

SDAVFs result in a congestive myelopathy with resultant neurological deficits.[5] As blood is shunted from the high-pressure arterial system to the inter-medullary venous plexus of the spinal cord, there is a reduction of the arterio-venous pressure gradient resulting in increased venous pressures and congestion of spinous veins. As a consequence of venous congestion, there is edema in the spinal cord. Furthermore, the congestion leads to decreased blood flow to the spine resulting in ischemic changes. If the ischemia is persistent and severe, irreversible cellular injury can occur.

The spinal cord edema and ischemia manifests clinically as neurological dysfunction with symptoms usually involving the lower extremity.[1,3,6] Symptoms typically begin with gait disturbances, paresthesias, and radicular pain. As the disease progresses, the motor and sensory disturbances become more severe. Additional symptoms can include bladder and bowel dysfunction, and erectile dysfunction. Three years following the onset of symptoms over 90% of patients were unable to walk.[3] It is imperative to treat patients early in their disease course to prevent the disabling symptoms of SDAVFs.

SDAVFs are delayed to diagnosis on average by 16 months.[3] Once diagnosed, the

treatment varies upon location and hospital preferences. Lesions typically are treated via endovascular techniques or surgical approach if the lesion is located near the spinal artery. These procedures are done under anesthesia to allow better visualization of the spinal artery, which depending upon location to the lesion, can be a contraindication to the endovascular approach. It also improves the ability to see residual fistula after embolization. Although open surgery may result in a higher cure rate than endovascular approach, the benefit to risk must be considered including longer recovery time, longer hospital stay, infection, and blood loss.

Our patient had a SDAVF arising from bilateral lateral sacral arteries with a four-year history of progressive bilateral lower extremity weakness. Over the next three days, our patient had marked interval improvement with strength returning to her lower extremities as well as improved control of bowel and bladder. Although rare, it is important to understand the presenting symptoms of SDAVF to prevent the long time to diagnosis as well as the fact that once diagnosed, via catheter-based embolization, it is a very treatable disease.

References

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