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Case Report

Spinal dural arteriovenous fistula: a case-based review

Gargee M. Pore*, Brig S. P. Gorthi

Department of Neurology, Bharati Hospital and Research Centre, Pune, Maharashtra, India

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***Correspondence:**
Dr. Gargee M. Pore,
E-mail: gargeepore@gmail.com

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ABSTRACT

Spinal dural arteriovenous fistula is an abnormal connection between radicular arteries and spinal venous plexus. This condition is relatively rare with annual incidence of five-ten cases per million. We present a case of a 62-year-old male with complaints of tingling in feet since six months and weakness in legs since 1 hour. Lower limb weakness progressed over a day to complete paralysis and bladder incontinence. Magnetic resonance imaging (MRI) spine showed a dural hypointensity over D6-D11 with abnormal vessels around spinal cord. Patient underwent spinal angiography which showed an intramedullary arteriovenous malformation causing compression. Arterial embolization was done within 24 hours from symptom onset and he ambulated within a week. AV malformations are a rare cause of compressive myelopathy presenting with acute paraplegia. Advances in endovascular techniques have made it possible to treat them early and prevent irreversible damage.

Keywords: Dural fistula, Paraplegia, Myelopathy

INTRODUCTION

Spinal dural arteriovenous fistulas (DAVFs) are the most common type of spinal vascular malformation, accounting for up to 85% of lesions with a male: female ratio of 5:1.[1] Majority of patients show a combination of gait difficulty, sensory disturbance, and involvement of sacral segments.[2] They are grossly underdiagnosed even in the era of neuroradiological advents, resulting in considerable morbidity. Spinal angiography is the gold standard in localizing vascular malformations and confirming degrees of arterial drainage outflow. Therapy is aimed at occluding the feeder vessel so as to arrest the progression of disease and improvement of symptoms.

CASE REPORT

A 62-year-old gentleman complained of tingling sensation in limbs for six months. One day he presented to the emergency department with an acute onset numbness and tingling in the hips and legs since two hours. He drove to work but was not able to walk from his car upon arrival due to weakness. This was accompanied by a short episode of 5/10 retrosternal chest pain and anxiety. Clinical examination did not reveal any long tract involvement, however neuroimaging showed features of long segment myelitis.

Past medical history included hypertension and hypothyroidism which was controlled with appropriate medications.

On physical examination, the patient was oriented and cooperative, his vital signs were normal. Muscle tone was absent in lower extremities. Deep tendon reflexes were diminished in both legs, and the abdominal and cremasteric reflexes were absent. His perianal sensation was absent as well. He had sensitive level at T10 below which all sensations were diminished. Patient had no tenderness over spine. He was fully paraplegic and had total urinary incontinence.

Non-contrast-enhanced magnetic resonance imaging (MRI) of the lumbar spine demonstrated the non-enhancing long segmental intramedullary hypointensity in the dorsal cord from D6-D11. Abnormal vessels around

cord surface showing post-contrast enhancement. Initial diagnostic impression was spinal cord infarct versus transverse myelitis. The patient was placed on steroids for spinal cord edema. The MRIs additionally revealed abnormally dilated vascular structures within thoracic and lumbar epidural space, consistent with SDAVF syndrome. Abnormal flow dynamics within the spinal cord secondary to the arteriovenous fistula was presumed to be causing cord ischemia and edema.

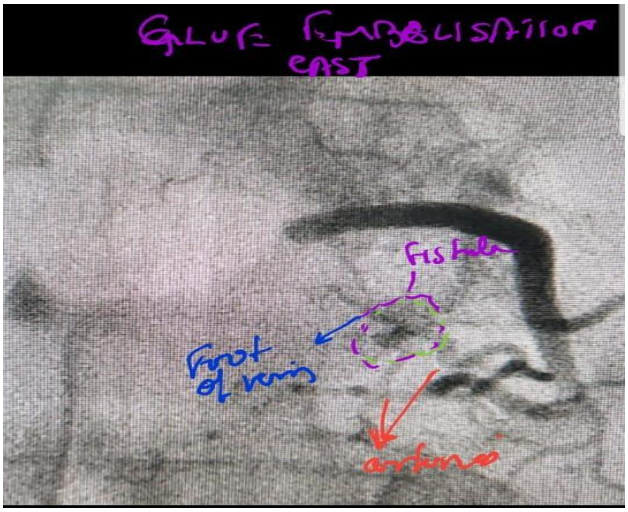

Figure 1: Frontal digital subtraction angiogram (during intervention) with both liquid embolization and microcoils (red arrow). Image during right L2 lumbar artery contrast injection shows abnormal filling of a dilated right radicular ascending vein (blue arrow) and dilated epidural veins.

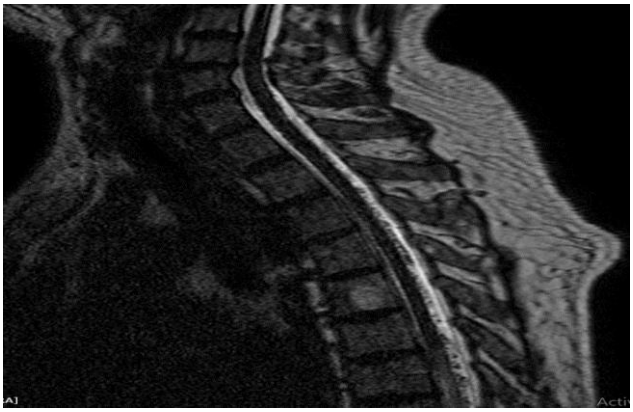

Figure 2: Long segment cord edema is seen from D6-7-disc level upto the tip of conus. 3D DRIVE sagittal images show abnormal mesh work of vessels along the cord surface in this segment as well as above it upto D2 level, which appears regressed compared to the pre-operative MRI scan.

Selective spinal angiography was initiated using a 4-French Shepherd Hook catheter, bilaterally. Normal spinal cord vascular anatomy was observed, including a radiculomedullary branch off the left T9 artery supplying the anterior spinal axis. Significant finding during angiography was an epidural fistula supplied by the right L1 and L2 arteries draining into the right L2 radicular veins.

Associated venous hypertension was demonstrated by slow filling of the spinal cord veins at the above spinal levels. Considering these findings, super-selective angiography and endovascular embolization with N-butyl cyanoacrylate and platinum microcoils of the leading L1 and L2 arteries were performed (Figures 1 and 2). Patient experienced immediate improvement in sensorimotor symptoms. He regained partial lower extremity muscle strength (grade 3/5), sensation, and bladder sphincter tone. A postoperative MRI demonstrated expected improvement in spinal cord edema as well as decreased abnormal spinal vascular engorgement.

DISCUSSION

This gentleman presented to the emergency department with nonspecific symptoms of paresthesia in hips and legs. Aforementioned symptoms might also be indicative of sciatic nerve compression, restless legs syndrome, peripheral diabetic neuropathy, chronic low back pain with radiculopathy, Guillain-Barre syndrome, or Sjogren's syndrome. The formation of blood vessels in the brain and spinal cord requires proliferation of endothelial cells that appears to be driven by the fork-head box O (FOXO1) transcription factor.

This inherited or sporadic vascular malformation presents in males in the fifth to sixth decade, with progressive paraplegia and associated bladder or bowel dysfunction. Lower extremity sensory disturbances are often patchy and ill-defined. The clinical course of SDAVFs is typically slowly progressive. However, the present case belongs to 5-10% of patients that experience an acute or a devastating neurological deterioration over the course of hours or days [3]. The severe neurological deterioration characterized by lower limb paralysis and bowel/bladder dysfunction due to a necrotizing myelopathy caused by a spinal vascular lesion is known as the Foix-Alajouanine syndrome.[4] Decreased muscle strength and spasticity is symptomatic of corticospinal tract impairment.

Structural MRI analyses with contrast-enhanced methods further buttressed the above neurologic finding as aberrant dilated and sprouting vessels within the spinal canal were visualized. Constructive interference in steady-state (CISS) complements conventional FSE sequences and better demonstrates tortuous and dilated venous structures.

Spontaneous closure of a SDAVF without treatment is extremely rare. Treatment options of a SDAVF include endovascular embolization and surgical ligation of the fistula. The success rate of surgery (98%) is higher than that of embolization (46%).[5] Following complete occlusion of the fistula, only two thirds of all patients have a regression of motor symptoms and one third show an

improvement of their sensory disturbances.[6] Impotence and sphincter disturbances are seldom reversible, and pain may persist.

CONCLUSION

AV malformations are a rare cause of compressive myelopathy presenting with acute paraplegia. Advances in endovascular techniques have made it possible to treat them early and prevent irreversible damage.

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