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Microsurgical intraluminal obliteration of type IV perimedullary arteriovenous fistula with an in situ hemostatic agent: illustrative case

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BACKGROUND Spinal arteriovenous fistulas (SAVFs) are underdiagnosed entities that can lead to severe morbidity from spinal cord dysfunction or hemorrhage. Treatment options include endovascular embolization or direct surgical obliteration at the level of the arteriovenous shunt. The authors present a case of intraluminal microsurgical access for occlusion with a hemostatic agent of a type IV SAVF near the conus medullaris as an alternative to clip occlusion to avoid nerve root compromise.

OBSERVATIONS Temporary microsurgical clipping of the SAVF led to nerve root compromise detected via intraoperative monitoring. Instead, the authors advanced elongated pieces of a hemostatic agent directly into the arterial lumen via arteriotomy to create direct obliteration of the fistula without intraoperative monitoring changes.

LESSONS In patients unable to tolerate clipping of the SAVF because of nerve root involvement and neurophysiological signal decline, open access of the vessels and direct intraluminal obliteration using a hemostatic agent should be considered as an alternative method of fistula occlusion.

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KEYWORDS spine; arteriovenous; fistula; angiography

Spinal arteriovenous fistula (SAVF) is a rare type of vascular malformation caused by an abnormal connection between the arteries and veins in the dural membrane surrounding the spinal cord. The initial work-up of SAVF can be challenging because of the nonspecific and insidious nature of symptoms, and diagnosis is usually delayed. Once the diagnosis is established, therapeutic options include endovascular embolization and open surgical obliteration of the fistula by excision, clipping, or ligation.[1–4]

Here, we report a case of type IV intradural perimedullary SAVF in which initial application of a temporary aneurysm clip was followed by the intraoperative loss of motor evoked potentials (MEPs) of the anal sphincter and bilateral feet. We opted for clip removal and fistula obliteration with the microsurgical intraluminal placement of hemostatic material.

Illustrative Case

History and Examination

A 60-year-old female presented with a long-standing history of back and bilateral lower extremity pain consistent with neurogenic claudication. Magnetic resonance imaging (MRI) revealed multilevel degenerative changes throughout the lumbar spine, which were not thought to be correlative with her symptoms. A prominent vascular structure in the lumbar cerebrospinal fluid spaces raised the possibility of an arteriovenous fistula or arteriovenous malformation (Fig. 1). The patient had years of increasing foot paresthesias, difficulty with ambulation, and retrospective reports of urinary incontinence. On motor examination, the extensor hallucis longus and anterior tibialis showed mild weakness (4+/5). Diagnostic spinal angiography showed a type IV SAVF near the conus medullaris. Arterialized flow from the artery of Adamkiewicz at the level of T11 traveled caudally to the L5–S1 level, then rostrally toward the conus, and then caudally again before draining into the epidural venous plexus (Fig. 2).

Surgery

An L3–S1 laminectomy was performed with the patient under general anesthesia with somatosensory evoked potential, MEP, and electromyography (EMG) monitoring. A midline dural incision was

ABBREVIATIONS EMG = electromyography; ICG = indocyanine green; MEP = motor evoked potential; MRI = magnetic resonance imaging; SAVF = spinal arteriovenous fistula.

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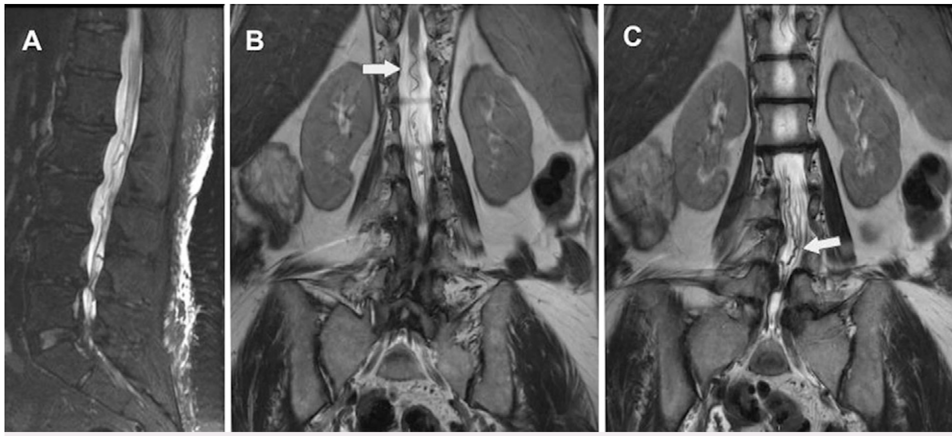

FIG. 1. Sagittal (**A**) and coronal (**B and C**) T2-weighted MRI showing abnormal serpentine vasculature at the level of the lumbar cistern (*white arrows*).

made, revealing the intradural fistula. Two separate serpentine vessels were densely adherent to one another. There was no clear dissection plane to separate the abnormal vasculature from surrounding nerve roots, which appeared to be splayed circumferentially around the fistula (Fig. 3). After meticulous dissection, a temporary aneurysm clip was placed on the presumed fistulous point. The mean arterial pressure was intentionally lowered to below systolic blood pressure of 90 mm Hg to observe for neurophysiological changes. This mean arterial challenge

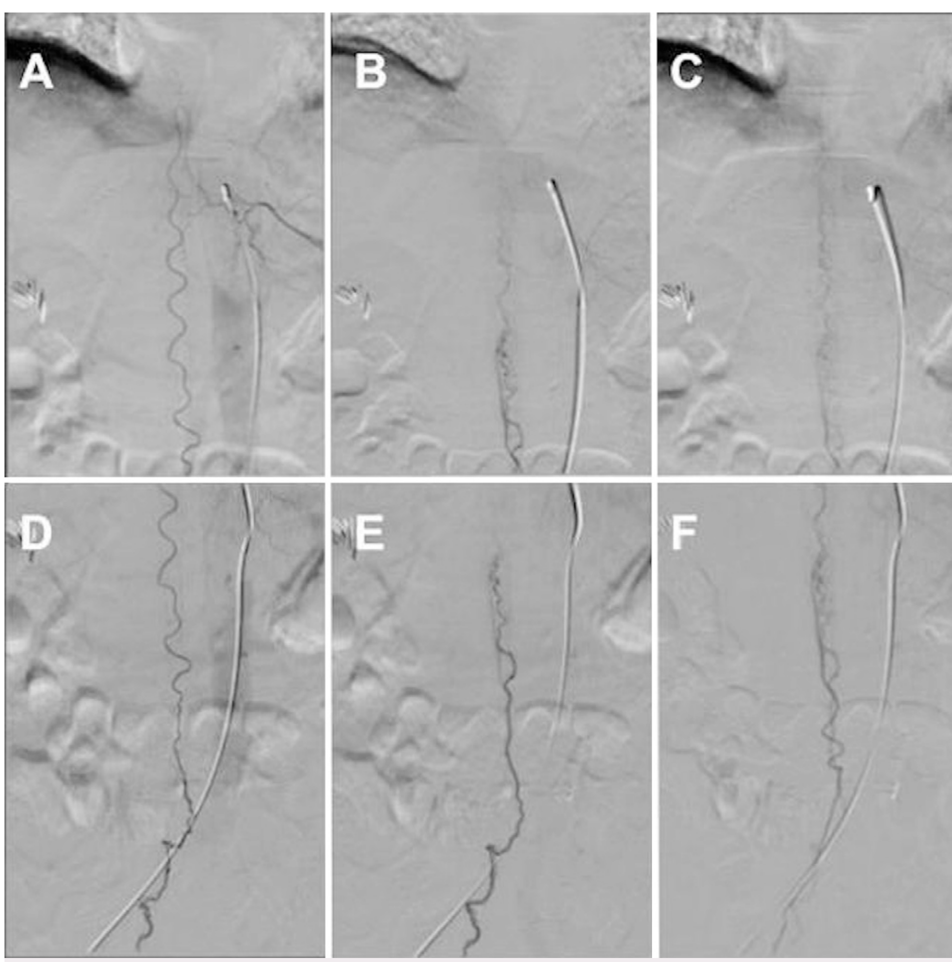

FIG. 2. Diagnostic angiography showing a type IV perimedullary arteriovenous fistula of the distal anterior spinal artery of Adamkiewicz. The flow traveled sequentially from the artery of Adamkiewicz caudally to L5–S1 (**A**, **D**), then turned rostrally toward the conus (**B**, **E**) and then caudally again (**C**, **F**).

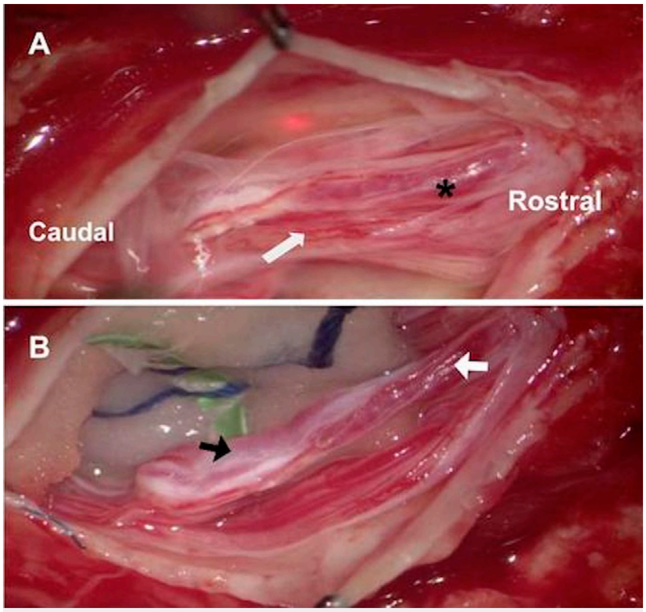

FIG. 3. A: *White arrow* indicates nerve roots tightly attached to the fistulous vessel (*asterisk*). **B:** *Black arrow* indicates an arterialized vessel with caudal flow. *White arrow* indicates an arterialized vessel with rostral flow.

maneuver was performed because preoperative angiography showed rostral blood flow toward the conus, and we wanted to ensure that any transient postoperative blood pressure changes would not result in conus ischemia. Also, indocyanine green (ICG) angiography was performed at that time with a quick and transient release of the temporary clip to observe the flow pattern. Interestingly, there was no flow within the fistula.

Five minutes after temporary clip placement, anal sphincter and bilateral feet MEP signals were absent. Signals returned to baseline levels after clip removal and blood pressure elevation. It was unclear whether ischemia or mechanical clip compression caused the electrophysiological dysfunction. EMG stimulation all around the fistula surface, including the area of temporary clipping, confirmed the presence and activation of the bilateral anal sphincter, gastrocnemius, and anterior tibialis rootlets. Further dissection did not allow greater separation of the fistula from the nervous tissue. ICG angiography was performed again without any aneurysm clips present. We observed arterialized ICG flow from rostral to caudal in one of the serpentine fistula vessels, followed by caudal to rostral arterialized flow in the second fistula vessel, followed by normal venous outflow in the surrounding vessels. Because of the above electrophysiological changes with application of the temporary clipping, the procedure was aborted for further discussion of the care plan with the patient. Postoperatively, the patient had normal motor and anal sphincter function. We discussed the risks of lower nerve root dysfunction with surgical fistula obliteration versus the natural history of hemorrhage of conus medullaris dysfunction, and the patient elected to undergo definitive surgical treatment.

A similar surgical approach was performed 6 days later. Given the loss of motor signal with temporary clipping, we obliterated the fistula with intraluminal packing to stop the flow. A longitudinal arteriotomy of the fistulous point parallel to the nerve rootlets was performed. The lumen was then packed with a piece of hemostatic oxidized cellulose. A figure-of-eight suture of the arterial wall was used to hold the cellulose. This process was repeated for the second fistula vessel. Afterward, the dura and incision were closed in a standard fashion (Fig. 4).

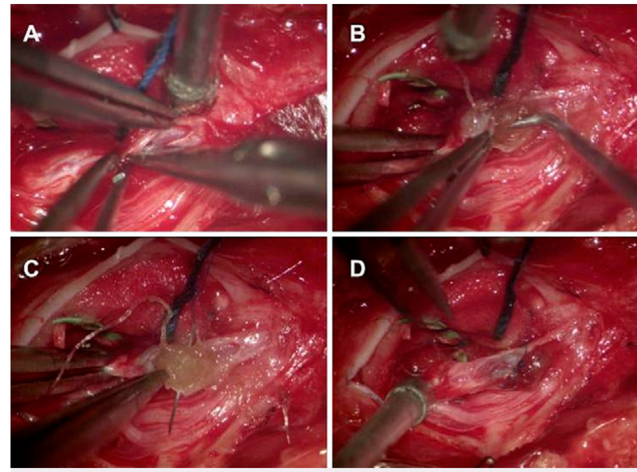

FIG. 4. Vessel obliteration. **A:** The assistant uses forceps for proximal and distal control during arteriotomy. **B:** Packing with the hemostatic agent in the lumen of the artery. **C:** Securing the hemostatic agent to the vessel wall. **D:** Final suturing of the arteriotomy.

Outcome

The patient had temporary urinary retention for a few days postoperatively, which eventually resolved. Bilateral lower extremity sensory and motor function were normal on bedside examination following surgery. Repeat angiography 1 month after surgery showed normal flow within the anterior spinal artery and obliteration of the fistula (Fig. 5).

Patient Informed Consent

The necessary patient informed consent was obtained in this study.

Discussion

Observations

SAVFs are the most common spinal vascular malformations, representing 70% of all vascular pathologies. There is an incidence of approximately 5–10 cases per 1 million persons per year, most commonly affecting the lower thoracic and upper lumbar area.[5–7] The overall incidence of type IV SAVF is unknown, but case series have reported a 17% rate of the total of dural SAVFs.[8]

The initial symptoms of SAVF are not specific and can include back pain with or without radiculopathy, paresthesias, myelopathy, weakness, loss of bowel and bladder control, or erectile dysfunction.[3,5,6] These can occur with variable acuity.

MRI is the most frequent test to prompt the diagnosis initially. The most common findings on MRI of thoracolumbar SAVF are enlargement of the distal spinal cord and conus with associated pial vascular engorgement and the presence of “flow voids” and cord edema on the T2 sequence.[5,7]

The gold standard diagnostic test is catheter-based spinal angiography. This provides dynamic diagnostic information regarding arterial inflow, fistula location, and venous outflow, enabling concurrent embolization

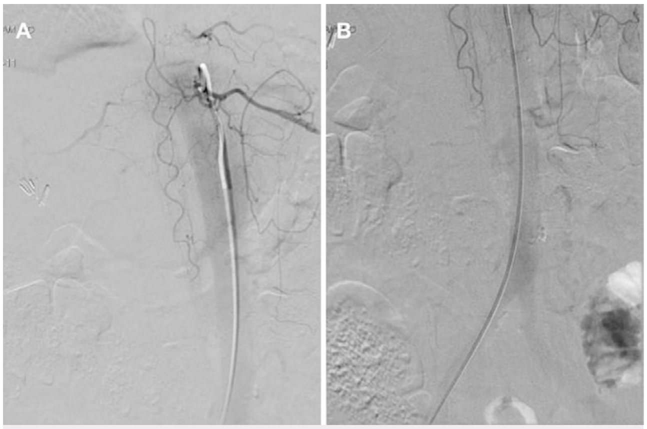

FIG. 5. A and B: Postoperative angiography shows the resolution and obliteration of type IV perimedullary arteriovenous fistula.

after diagnosis in the right setting.[7] The technique should be focused on assessing the intercostal branch, radiculomedullary artery, anterior spinal artery, and the artery of Adamkiewicz.

Multiple classifications have been proposed regarding spinal vascular malformations, but there has been no consensus about the most optimal assessment tool. This report focuses on type IV intradural perimedullary SAVF, first described by Djindjian et al.[9] among their series of 150 patients but later defined as such by Heros et al.[10] Type IV is most commonly considered a slow-flow fistula with moderate venous enlargement, supplied by either the anterior or posterior spinal arteries, and is frequently located in the ventral surface of the conus medullaris or filum terminale, presenting with progressive radiculopathy or subarachnoid hemorrhage.[11]

Multiple methods can be used to obliterate the fistulous connection surgically. These can include cauterization and sectioning of the fistulous point, resection of the fistula, application of temporary clips causing clotting of the vessels, or the use of permanent small hemoclips to interrupt the flow.[2,4,12–18]

Despite the constant improvements in endovascular technique, several studies on microsurgical approaches have demonstrated lower rates of treatment failure and higher rates of symptom resolution in treating SAVF.[1,13,19–21] Goyal et al.[19] reported that microsurgery was associated with significantly lower odds of initial treatment failure and late recurrence and high odds of clinical improvement compared with endovascular embolization. Koch et al.[1] reported on their 10 years of institutional experience treating SAVFs. They found that 100% of patients who had undergone microsurgical clipping exhibited complete occlusion compared with 65% of patients in the embolization group. Additionally, 93% of patients had symptom resolution compared with 88% in the microsurgical and endovascular groups. Rothrock et al.[12] reported a definitive solution for refractory SAVF by microsurgical ligation in patients who had undergone initial embolization that failed to resolve the shunt. On the other hand, Sure et al.[22] favored an interdisciplinary and combined approach with an initial endovascular technique, followed by microsurgical techniques. This experience was demonstrated in a pediatric case series of type IV SAVFs. Specifically addressing the microsurgical treatment of type IV SAVF, Barrow et al.[11] described better outcomes by placing an aneurysm clip across the fistula to minimize bipolar coagulation near the spinal cord.

The benefits of using intraoperative monitoring for basic safety measurements have been demonstrated in the literature on the spinal cord, filum terminale, and conus levels.[23–26] The common factor in surgical techniques is flow occlusion via a transverse mechanical force against the vessel wall. On the basis of our neurophysiological recording data, this occlusion technique likely compromised nerve root function, given that separation of the fistula and nerve roots with a microsurgical technique did not seem possible.

In our case, placing a temporary clip on the fistula and dropping blood pressure with subsequent MEP loss suggests more clipping effect on the bilateral rootlets attached to the SAVF. Entrapment of the rootless with the clip decreases functions much more slowly because it involves the occlusion of vascular structures and behaves like a cushion so the neural axonal blockage does not manifest immediately.

The preoperative angiogram showed flow caudally from the conus to L5–S1, then rostrally to the conus, and then caudally again. We induced hypotension intraoperatively as a challenge maneuver to help predict if the elimination of rostral blood flow and possible unplanned episodes of postoperative hypotension would lead to conus ischemia.

Our goal in the intraluminal application of a hemostatic agent was to decelerate the flow of the fistula and allow gradual obliteration without the direct external transverse compressive effect of the fistula that seemed to place adjacent deeply attached nerve roots at risk of injury.

Before opting for a microsurgical technique, the endovascular approach was deemed dangerous to perform. In order to reach the fistulous point, the access had to be done through the tortuous anterior spinal artery. Navigating and embolizing the intradural fistula safely was not possible, because it was too far from the desired fistulous point.

Lessons

Microsurgical intraluminal occlusion of fistulous vessels using a hemostatic agent is feasible when direct external compression with clips or resection appears to cause significant nerve root dysfunction due to its closed anatomical relationship and inability to create a safe surgical plane. The decision to use this technique can be based on intraoperative anatomical and dissection findings in conjunction with objective neurophysiological changes.

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Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: Lara-Reyna, Klopfenstein. Acquisition of data: Lara-Reyna, Garst. Analysis and interpretation of data: Lara-Reyna. Drafting the article: Lara-Reyna, Garst, Winslow. Critically revising the article: Lara-Reyna, Winslow, Klopfenstein. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Lara-Reyna. Administrative/technical/material support: Lara-Reyna, Garst. Study supervision: Lara-Reyna, Klopfenstein.

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