CASE REPORT

Angiographic suppression of the artery of Adamkiewicz by venous hypertension resolving after embolization in a case of spinal epidural arteriovenous fistula

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SUMMARY

A case of complete angiographic suppression of the artery of Adamkiewicz and anterior spinal artery in a patient with a spinal epidural arteriovenous fistula (AVF) is reported. Slow flow AVFs typically present with progressive myelopathy secondary to spinal venous hypertension (SVH). The lack of a normal venous phase during angiography and its restoration after treatment is commonly observed with these lesions, yet a similar phenomenon seems exceptional at the arterial level. Right T11 intercostal artery angiograms obtained before and after treatment of a left L4 epidural AVF documented the initial suppression of the artery of Adamkiewicz and anterior spinal artery, and their normal appearance immediately after correction of the SVH by embolization. This report confirms that SVH can angiographically suppress prominent and functionally important spinal arteries, re-emphasizing the potential role played by secondary arterial changes in SVH induced myelopathy. This hemodynamic phenomenon also represents a potential pitfall during diagnostic and therapeutic endovascular procedures.

BACKGROUND

Aminoff *et al*¹ suggested in 1974 that the progressive myelopathy typically associated with slow flow spinal arteriovenous fistulas (AVF), such as dural and epidural AVFs, was the consequence of spinal venous hypertension (SVH). The absence of a normal venous phase during spinal angiography in two cases of spinal AVFs reported by Launay *et al*² in 1979 was consistent with this hypothesis. Merland *et al*³ emphasized 1 year later that concomitant impairment of spinal venous drainage was necessary for these low pressure and slow flow lesions to cause spinal cord damage.

SVH generally prevents the angiographic visualization of the perimedullary venous system. In fact, documentation of a normal venous phase during spinal angiography is considered a strong sign against a diagnosis of spinal AVF.⁴ SVH is also believed to impair the spinal arterial circulation. We describe here a case of spinal epidural AVF, in which the artery of Adamkiewicz and the anterior spinal artery, not visible during initial diagnostic angiography, were only identified after eradication of the arteriovenous shunt and correction of the SVH.

CASE PRESENTATION

A 48-year-old man presented with rapidly progressing paraparesis. CSF analysis showed mild pleocytosis and elevated proteins (127 mg/dL). MRI of the thoracic and lumbar spine revealed abnormal signal from T9 to the conus medullaris. Intravenous steroids were administered for suspected transverse myelitis, without significant improvement. The onset of perineal numbness and sphincter dysfunction prompted a repeat course of steroids that remained ineffective. The patient was referred to our institution 8 months later, after MRI documented disease progression and prominent perimedullary vessels (figure 1A). At admission, the examination demonstrated an L1 sensory level, flaccid paraplegia, an atonic anal sphincter, and patellar and Achilles areflexia.

INVESTIGATIONS

Spinal angiography confirmed the presence of a left L4 spinal epidural AVF with a relatively high flow shunt between a radiculomeningeal artery and an isolated pouch of epidural plexus draining into a prominent radiculomedullary vein (figure 1B, C). The artery of Adamkiewicz and the anterior spinal artery, not visible during the diagnostic angiogram, were readily identified after endovascular treatment of the AVF and correction of the SVH (figure 1D, E). The only difference between the two studies performed 1 day apart in the same angiography suite by the same operator was the use of conscious sedation during diagnostic angiography and general anesthesia during embolization. The selective injection technique (handheld syringe injection), contrast dilution (75% contrast/25% saline), and angiography protocols (variable frame rate protocol, 2.4 μ Gy/fr) were identical.

DISCUSSION

The angioarchitecture of spinal dural AVFs was first appreciated by Kendall and Logue in 1977.⁵ The separate existence of spinal epidural AVFs has been recognized for almost as long.³ Dural and epidural AVFs typically present with a progressive myelopathy induced by SVH,¹ a concept confirmed by the intraoperative pressure measurements performed in 1989 by Hassler *et al*.⁶ Spinal AVFs also impair the spinal arterial circulation.^{2,3} For Willinsky *et al*,⁴ the observation of a delayed spinal circulation strongly suggests the presence of an AVF. Yet,

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Figure 1 A 48-year-old man with progressive myelopathy. (A) MRI, STIR, sagittal view of the thoracic and lumbar spine, showing extensive signal anomaly from the conus medullaris up to the level of T4; prominent 'flow voids' indicate enlarged perimedullary venous structures. (B) DSA, left L4 injection, longitudinally combining two acquisitions; a prominent left L4 radiculomedullary vein drains the arteriovenous fistula (AVF) into the perimedullary venous system, which is opacified up to C2 (top not shown). Note some degree of antegrade drainage towards the epidural plexus at L2, T3, and C7. Overall, the paucity of functionally significant radiculomedullary veins is consistent with the concept of diffuse venous disease underlying the formation of spinal AVFs and the development of a venous hypertensive myelopathy. The black arrow in Figure 1B marks the left L4 radiculomedullary vein. (C) DSA, superselective injection of a left L4 radiculomedullary branch (small white arrows) that vascularizes the spinal epidural AVF. The small black arrow corresponds to the microcatheter tip position. The large black arrow points at the arteriovenous shunt itself, while the large white arrow indicates the terminal segment of the left L4 radiculomedullary vein, just proximal to its connection with the epidural pouch. The feeding artery, the epidural pouch, and the proximal segment of the radiculomedullary vein were embolized with n-butyl cyanoacrylate glue. (D) DSA, selective injection of the right T11 intersegmental artery (ISA) (diagnostic angiography prior to embolization); no spinal contribution is noted. (E) DSA, selective injection of the right T11 ISA (control angiography immediately after embolization); the artery of Adamkiewicz and the anterior spinal axis are well opacified. The injection technique (hand injection), contrast dilution (75% contrast/25% saline), the angiography suite, and the operator were identical.

complete angiographic suppression of a significant radiculomedullary artery has, to our knowledge, rarely been emphasized before. In a recently published report, a prominent left T4 anterior radiculomedullary artery only became angiographically detectable after treatment of a right T4 SEAVF.⁷ To explain this finding, a change in either the patient's breathing pattern (conscious sedation vs general anesthesia) or in the hemodynamic conditions of the spinal circulation after eradication of the fistula was postulated. While similar reasoning could be applied to our case, the larger size of the artery of Adamkiewicz renders a difference in breathing pattern an unlikely explanation for its non-visualization prior to treatment. We suspect that SVH in our patient was sufficient to slow the arterial circulation beyond an angiographically detectable threshold, and that correcting the hemodynamic disturbance by embolizing the AVF restored both normal arterial flow and angiographic conspicuity. This assumption is consistent with the intraoperative documentation of increased spinal arterial flow velocities after the treatment of dural AVFs.⁶

While restoration of an initially absent venous phase is commonly observed in patients with treated spinal AVFs, a similar phenomenon seems exceptional at the arterial level. A higher degree of SVH is likely required to prevent the angiographic detection of a significant radiculomedullary artery. In this regard, it is important to note the slightly atypical angioarchitecture of our patient's AVF, with a prominent feeding artery transitioning almost seamlessly into an epidural venous pouch connected to a large radiculomedullary vein. In their hemodynamic study of spinal AVFs, Hassler⁸ noted that the pressure drop was highest at the shunt site itself, "due to the abrupt change in the diameter of the whole vessel" where "small caliber meningoradicular feeders emptied into the larger draining vein". The absence of significant caliber transition in our

patient probably resulted in a higher venous pressure compared with more typical, intricate, and slower spinal AVFs.

It was initially feared that the non-visualization of the spinal arterial system carried negative prognostic implications for our patient, suggesting, for example, diffuse spinal ischemia from arterial thrombosis in addition to the SVH induced myelopathy. The immediate documentation of an unremarkable arterial circulation after embolization was welcomed but its impact on long term functional outcome remains unknown.

This case report helps re-emphasize the potential role of secondary arterial disturbances in the clinical presentation of spinal AVFs associated with SVH, as suggested by Merland *et al* in 1980.³ We suspect that complex hemodynamic situations with compromised spinal cord arterial supply may result in sustained ischemic injury and lead to less satisfying clinical outcomes.

Finally, the possible existence of non-documented radiculomedullary arteries should always keep angiographers vigilant during the endovascular treatment of spinal AVFs with significant SVH, particularly in the presence of larger and faster

Learning points

- ▶ Spinal venous hypertension (SVH) induced by slow flow spinal arteriovenous fistulas (AVFs) can prevent the angiographic detection of functionally important spinal arteries.
- ▶ Secondary arterial flow impairment may represent a negative prognostic factor for the functional outcome of patients treated for slow flow spinal AVFs.
- ▶ Angiographic suppression of important spinal arteries by SVH represents a pitfall during diagnostic and therapeutic spinal angiography.

arteriovenous shunts. Undetectable spinal contributors may originate from neighboring intersegmental arteries,⁷ or even from the intersegmental artery vascularizing the AVF itself, carrying in this case a significant risk of procedural complications.⁹

Competing interests None.

Patient consent Obtained.

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