

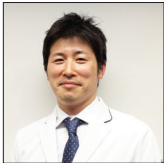


Case Report

Sacral arteriovenous fistula with lower thoracic cord edema without perimedullary vein enlargement

Motonori Ishii, Yusuke Nishimura, Yoshitaka Nagashima, Takafumi Tanei^{ORCID}, Masahiro Nishihori^{ORCID}, Takashi Izumi, Ryuta Saito
Department of Neurosurgery, Nagoya University School of Medicine, Nagoya, Aichi, Japan.

E-mail: Motonori Ishii - mottoii99@gmail.com; *Yusuke Nishimura - yusuken0411@med.nagoya-u.ac.jp; Yoshitaka Nagashima - nagashima4251@gmail.com; Takafumi Tanei - nsgtakasyun@msn.com; Masahiro Nishihori - nishihori@med.nagoya-u.ac.jp; Takashi Izumi - my-yuzu@med.nagoya-u.ac.jp; Ryuta Saito - ryuta@med.nagoya-u.ac.jp



*Corresponding author:

Yusuke Nishimura,
Department of Neurosurgery,
Nagoya University School of
Medicine, Nagoya, Aichi, Japan.
yusuken0411@med.nagoya-u.
ac.jp

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ABSTRACT

Background: Sacral dural arteriovenous fistulas (AVFs) are often undiagnosed at the initial presentation due to their rarity.

Case Description: For 1 year, a 71-year-old man developed progressive motor and sensory disturbances in both legs. Magnetic resonance imaging showed spinal cord edema with mild contrast enhancement at the T9–10 and T12 levels. Although mild venous dilatation was observed only at the cauda equina level, it was not initially recognized as abnormal. Blood and cerebrospinal fluid tests and spinal angiography of the lower thoracic to upper lumbar levels were nonspecific. The patient was unsuccessfully treated with three courses of high-dose intravenous methylprednisolone. Ultimately, following repeat spinal angiography (i.e., including the bilateral internal iliac arteries) that revealed a low-flow sacral dural AVF supplied by the right lateral sacral artery, the patient underwent successful surgical venous AVF occlusion/transection.

Conclusion: In cases of spinal cord edema without perimedullary abnormal flow voids, careful spinal angiography including the sacral spine is necessary even if only minimal venous dilation is initially observed at the cauda equina level.

Keywords: Magnetic resonance imaging, Sacral dural arteriovenous fistula, Spinal dural arteriovenous fistula

INTRODUCTION

Sacral dural arteriovenous fistulas (AVFs) comprise only 4–9% of all spinal AVFs.^[2,3] Patients usually present with progressive myelopathy attributed to venous hypertension. The definitive diagnosis of a sacral dural AVF may be established based on the presence of an early dilated draining vein seen on selective angiography accompanied by spinal cord edema, and magnetic resonance (MR)-documented intradural flow voids. However, this diagnosis is often missed as sacral AVFs are far removed from the site or edema in the lower thoracic spinal cord. Diagnosis of AVFs is best established utilizing spinal angiography including the internal iliac, median sacral, or iliolumbar arteries. Once identified, these lesions may be appropriately surgically treated.

CASE REPORT

A 71-year-old male presented with 3 months of progressive bilateral lower extremity motor weakness and dysesthesia. The spinal MRI showed two separate levels of spinal cord edema

at the T9–10 and T12 levels that were slightly enhanced with contrast-enhanced [Figures 1a-c]. Spinal angiography, including the aorta and segmental arteries from the T8 to L3, showed no abnormal shunting/flow. Three courses of high-dose intravenous methylprednisolone were unsuccessful. Nine months later, he presented with diffuse lower extremity hyperreflexia (4+) and underwent lumbar

spinal computed tomography angiography (CTA). The CTA documented a dilated vessel extending from the right first sacral foramen to the conus medullaris [Figure 1d]. Repeated spinal angiography, including bilateral internal iliac arteries and segmental arteries from T8 to L5, revealed a sacral dural AVF with a “shunt point” on the right involving the S1 root (i.e., supplied by the right lateral

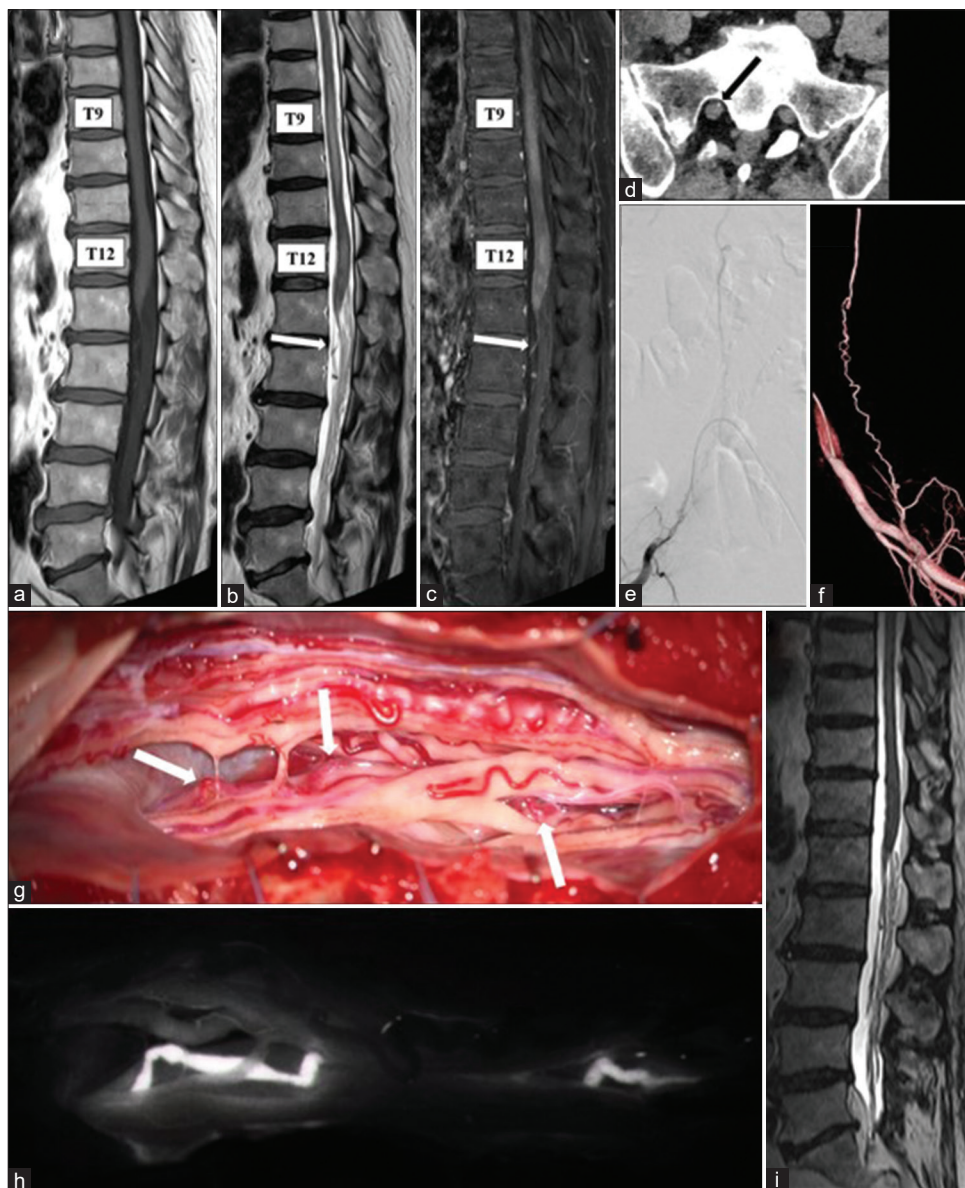


Figure 1: (a) T1-weighted sequence, (b) T2-weighted sequence, and (c) post-gadolinium T1-weighted sequence – initial magnetic resonance imaging at a local hospital, showing intramedullary edema at T9–10 and T12 levels and a mildly dilated vessel at the cauda equina level (white arrow) without perimedullary vein enlargement. (d) Contrast-enhanced computed tomography image at our hospital, demonstrating a dilated vessel in the right first sacral foramen (black arrow). (e and f) Right internal iliac artery angiograms, showing sacral arteriovenous fistula supplied by the right lateral sacral artery. (g and h) Intraoperative photos showing an arterialized vein (white arrows) along the S1 nerve root visualized early by the indocyanine green without other shunt vessels. (i) Magnetic resonance imaging 5 months after surgery, showing extinguished intramedullary edema.

sacral artery) [Figures 1e and f]. The slow flow passage of the intradural draining and right S1 radiculomedullary vein was consistent with a low-flow AVF.

Surgery

Following an L4–S1 laminectomy, intraoperative indocyanine green angiography revealed an arterialized “red vein” along the right S1 nerve root; no other shunting vessels were detected intradurally [Figures 1g and h]. The vein was successfully coagulated and severed; the pathology confirmed that it was a vein. Postoperatively, although numbness persisted, the patient’s ambulation and leg pain dramatically improved. Notably, the follow-up MR 4 months after surgery showed full resolution of the previously noted T9–10 and T12 cord edema [Figure 1i].

DISCUSSION

Even in the absence of obvious dilation of the perimedullary veins, sacral dural AVFs should be suspected in cases in which only mildly dilated veins of the cauda equina coexist with edema of the lower thoracic spine. MR findings associated with spinal dural AVFs include spinal cord edema, intramedullary contrast-enhancement, and perimedullary flow voids (i.e., highly suggestive of spinal dural AVFs/present in 85–88% of sacral dural AVFs).^[1-3] All 48 previously reported cases of sacral dural AVFs without additional spinal pathologies demonstrated abnormally enlarged perimedullary veins. Enlargement of the filum terminale veins is also seen in 75–100% of sacral AVFs.^[1-3] The previous reports have exclusively described the dilated veins in the cauda equina as the filum terminale veins, which are the only longitudinal collector veins seen below L2. Low-flow sacral dural AVFs may direct shunt flow exclusively to the anterior median spinal veins rather than the filum terminale veins through the radiculomedullary veins due to the shallow angle of inlet flow. The lightly dilated anterior median spinal veins may not be visualized as enlarged due to their location subpially.^[4] The dilatation of veins in the cauda equina, whether the filum terminale veins or radiculomedullary veins, is consistent with the presence of spinal dural AVFs. Thus, the combination of spinal cord edema and flow voids in the cauda equina facilitates the detection of sacral AVFs even without perimedullary flow voids.

CONCLUSION

A sacral dural AVF went undiagnosed for over a year in a 71-year-old male. Ultimately, a laminectomy from L4 to S1 allowed for the occlusion/resection of the arterialized AVF vein along the right S1 nerve root.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of Artificial Intelligence (AI)-Assisted Technology for assisting in the writing or editing of the manuscript and no images were manipulated using the AI.

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