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Keywords

Spinal epidural arteriovenous fistula, Transvenous embolization, Intramedullary hemorrhage, Hematomyelia

Highlights

- A hemorrhagic presentation of spinal epidural arteriovenous fistula (SEDAVF) is rare and an intramedullary hemorrhagic presentation has been reported in only two cases.
- We report a case of lumbar SEDAVF presenting with remote hemorrhage in the posterior column of the thoracic spinal cord.
- Unlike the good prognosis after treatment for patients with SEDAVF, our patient did not achieve independence because of residual loss of proprioception and vibration sense which were sequelae of remote spinal cord hemorrhage.

Full article title

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Introduction

A spinal epidural arteriovenous fistula (SEDAVF) is a rare spinal vascular malformation consisting of abnormal connections between the dorsal somatic artery and ventral epidural venous plexus. Most patients present with congestive myelopathy due to intradural perimedullary venous drainage, and others present with compressive symptoms because of engorged extradural venous drainage [1]. A hemorrhagic presentation of SEDAVF is extremely rare; to the best of our knowledge, an intramedullary hemorrhagic presentation has been reported in only two cases [2, 3], and remote spinal cord hemorrhage has not been reported to date.

We herein report a case of lumbar SEDAVF presenting with remote hemorrhage in the posterior column of the thoracic spinal cord. The patient was successfully treated by transvenous embolization (TVE); however, she did not achieve independence because of residual loss of proprioception and vibration sense which were sequelae of remote spinal cord hemorrhage.

Case presentation

An 82-year-old woman presented with an 8-month history of slowly progressive paraparesis and bilateral lower limb paresthesia. There was no history of spinal surgery or trauma. She was referred to our hospital because of sudden-onset ataxic gait due to impairment of proprioception and vibration sense. On admission, she was able to stand with assistance but unable to walk because of balance impairment and had impaired bladder function and occasional incontinence. Sagittal T1- and T2-weighted magnetic resonance imaging (MRI) showed spinal cord hemorrhage at the level of the eighth thoracic vertebra (Fig. 1a–c). Multiplanar reconstruction images of computed tomography angiograms in the early and late phases showed abnormal opacification of the ventral epidural venous

plexus at the level of the fifth lumbar vertebra with enlarged perimedullary veins (Fig. 1d, e). Imaging revealed no abnormal opacification, such as a varix, in the draining vein at the level of the eighth thoracic vertebra. An axial T2-weighted image at the level of the eighth thoracic vertebra showed low signal intensity in the posterior column (Fig. 1f). An axial maximum intensity projection image of the fifth lumbar vertebra showed abnormal opacification of the ventral epidural venous plexus (Fig. 1g). Digital subtraction angiography of the left fourth lumbar artery showed that the ventral epidural venous plexus was fed by the left fourth and fifth dorsal somatic branches (Fig. 2a). A late arterial-phase image showed the left fifth radiculoemissary vein draining into the left ascending lumbar vein associated with the ascending intradural perimedullary drainage (Fig. 2b). Anteroposterior three-dimensional digital subtraction angiography showed the angioarchitecture of the SEDAVF draining into the left common iliac vein through the left ascending lumbar vein (Fig. 2c). We treated the patient by TVE with coils via the transfemoral approach under general anesthesia. The tip of a microcatheter was advanced into the epidural venous pouch through the left fifth radiculoemissary vein supported by a 4- and 6-French coaxial guiding system, and TVE was then successfully performed (Fig. 2d, e). The final left fourth lumbar angiogram showed complete obliteration of the shunt (Fig. 2f). No procedure-related complications occurred. The patient's postoperative course was uneventful, and her gait disturbance gradually improved. Follow-up MRI showed disappearance of the signs of venous congestion (Fig. 2g). At the 12-month follow-up, the patient was ambulatory but required two canes because of loss of proprioception and vibration sense.

Discussion

SEDAVF is considered a relatively low-flow shunt exhibiting variable venous drainage patterns, such as intradural and epidural routes [4]. The clinical manifestations depend on the patterns of drainage and can range from congestive myelopathy to compressive myelopathy or radiculopathy resulting from an enlarged epidural venous plexus [4]. In a meta-analysis of 101 patients with SEDAV, a hemorrhagic presentation was observed in 10 patients (10%), including 5 patients with an epidural hematoma and 5 with subarachnoid hemorrhage [5]. No cases of

intramedullary hemorrhage were reported in that study. A more recent review described 17 cases of SEDAVF with a hemorrhagic presentation, including 11 patients with an epidural hematoma, 3 with subarachnoid hemorrhage, and 2 with spinal cord hemorrhage [2]. The causes of intramedullary hemorrhage in the latter two patients were explained by the venous congestion appeared in the vicinity of the SEDAVF (i.e., cervical and lumbosacral). To the best of our knowledge, ours is the first reported case of lumbar SEDAVF associated with spinal cord hemorrhage remotely appeared in the thoracic spine. In cases of spinal dural arteriovenous fistulas, an intramedullary hemorrhagic presentation has been reported; most of these patients had venous varices of the draining veins, which were thought to be the source of the hemorrhage [2]. However, in our patient (Fig. 1d, e) and two previously described patients, no venous varix was found on diagnostic angiography. Although the etiology of the hemorrhagic presentation remains unclear and the co-occurrence of two different conditions cannot excluded, we believe that the remote spinal cord hemorrhage in the middle of the T2 high signal intensity (from the middle of fifth to lower of eleventh thoracic vertebrae), which suggested venous congestion, might have resulted in the intramedullary hemorrhage in our patient.

There is a strong correlation between the presence or absence of symptoms in SEDAVF and the content of symptoms, depending on the types of venous drainage and the extent of venous congestion. After treatment, many of patients reported improvement of symptom correlated with the improvement of MRI findings. Our patient underwent successful treatment with TVE, resulting in complete obliteration of the lesion, and noted marked improvement in motor symptom. Follow-up MRI showed complete disappearance of spinal cord edema; however, she did not become independent because of residual loss of proprioception and vibration sense which were considered to be a sequela of spinal cord hemorrhage.

Conclusion

lumbar SEDAVF associated with remote spinal cord hemorrhage is an extremely rare condition caused by venous congestion distant from the vicinity of the arteriovenous fistula. In patients with an intramedullary hemorrhagic presentation, residual neurological deficits cannot be avoided even with successful treatment.

Disclosure of interest

The authors declare that they have no competing interest.

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Ethical approval

The patients were able to review the article and give their consent for publication. The patients were treated according to the current standards of care. This article was conducted in accordance with the Declaration of Helsinki from 1964 or any further amendment.

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Figure captions

Fig. 1. Sagittal (a) T1- and (b) T2-weighted magnetic resonance images of the thoracic spine obtained at a local community hospital show spinal cord hemorrhage at the level of the eighth thoracic vertebra (white arrow). (c) A sagittal T2-weighted image of the lumbar spine shows flow void signals. Note the hemorrhage (white arrow) of the thoracic spine. Multiplanar reconstruction images of computed tomography angiograms of the whole spine in the (d) early and (e) late phases show abnormal opacification of the ventral epidural venous plexus (black arrow) at the level of the fifth lumbar vertebra with enlarged medullary veins. Imaging shows no abnormal opacification, such as a varix, of the draining vein at the level of the eighth thoracic vertebra (black blank arrowhead). (f) An axial T2-weighted image at the level of the eighth thoracic vertebra shows abnormal low signal intensity (white arrow) involving the posterior column of the spinal cord. (g) An axial maximum intensity projection image of the fifth lumbar vertebra shows abnormal opacification (black arrow) of the ventral epidural venous plexus.

Fig. 2. (a) A left fourth lumbar artery digital subtraction angiogram shows abnormal opacification of the ventral epidural venous plexus (asterisk) fed by the left fourth (black arrows) and fifth (white arrows) dorsal somatic branches. (b) Another view in the late arterial phase shows the left fifth radiculoemissary vein (black arrows) draining into the left ascending lumbar vein (white arrowheads). Note the ascending intradural perimedullary drainage (white arrows). (c) An anteroposterior view of a three-dimensional digital subtraction angiogram shows the angioarchitecture of the epidural arteriovenous fistula draining into the left common iliac vein through the left ascending lumbar vein (white arrowheads). Unsubtracted images during embolization show the (d) first and (e) final coil configuration placed at the epidural venous pouch and the origin of the right fifth radiculoemissary vein. Note the microcatheter advanced through the left fifth radiculoemissary vein supported by a 4- and 6-French coaxial guiding system. (f) Final left fourth lumbar angiogram shows complete obliteration of the shunt. (g) Sagittal T2-weighted image obtained 3 months after embolization shows complete disappearance of the intramedullary hyperintensity signal.

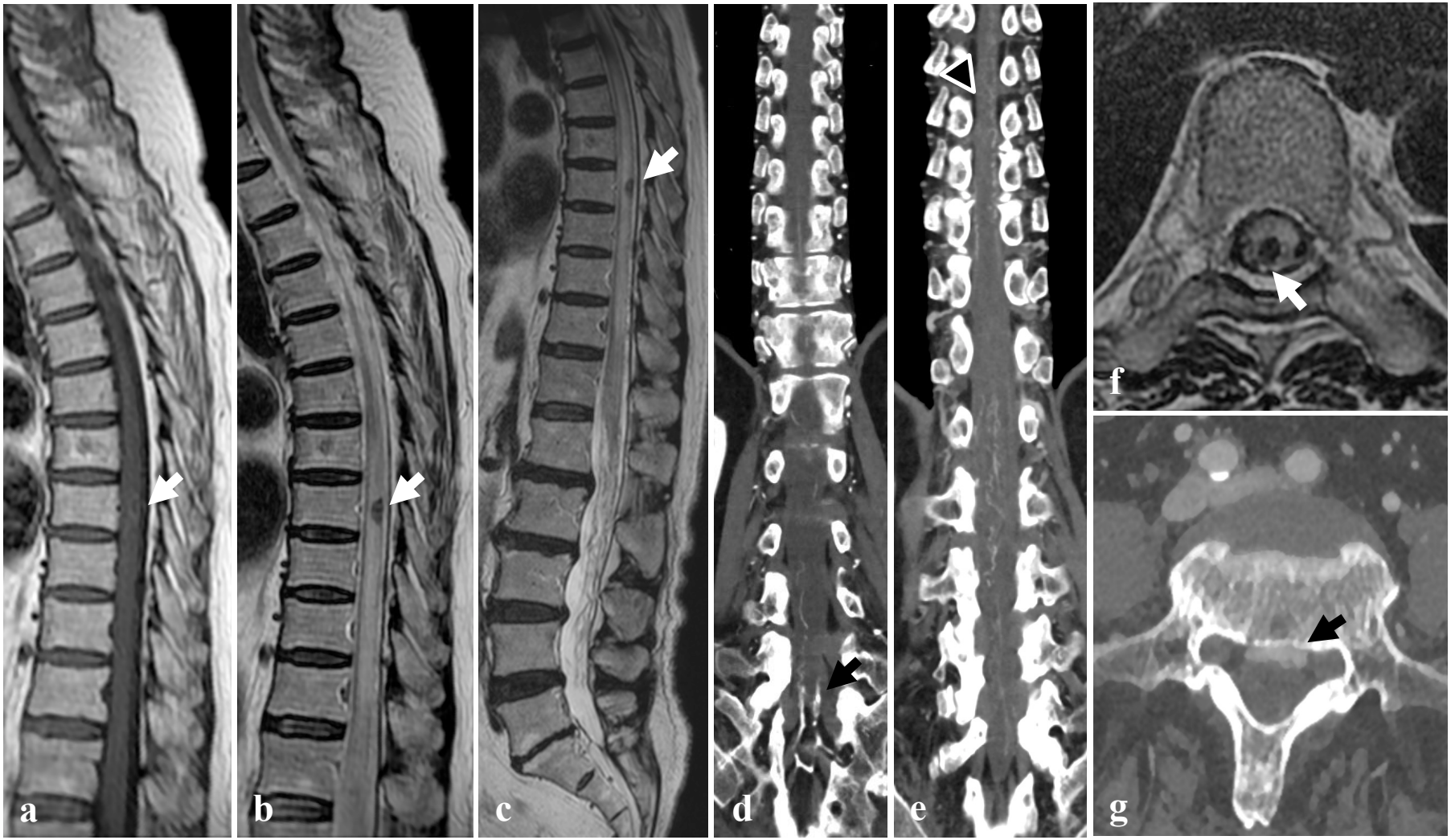

Fig. 1

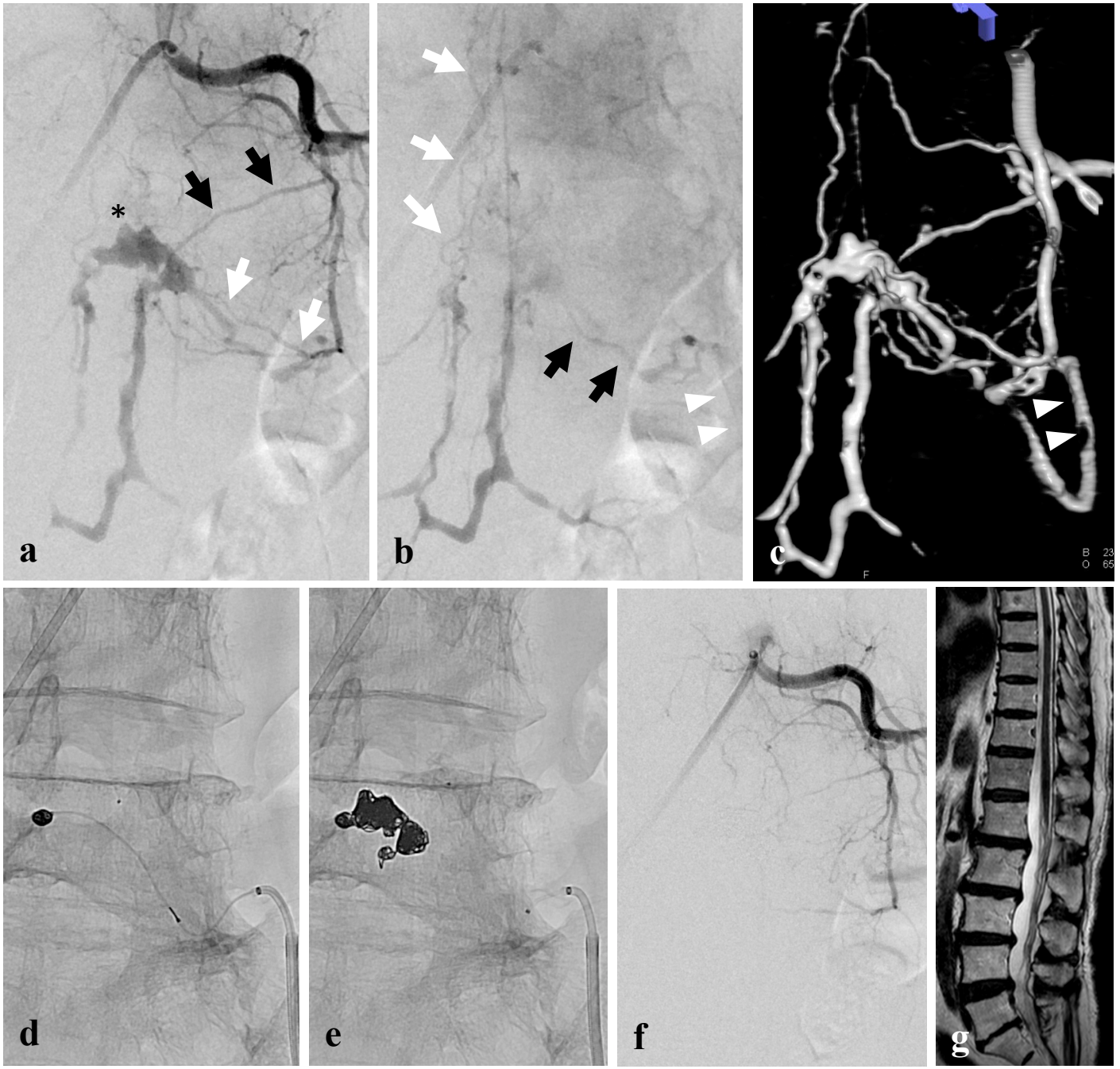

Fig. 2