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CASE REPORT

A case of arteriovenous fistula of the cauda equina fed by the proximal radicular artery: anatomical features and treatment precautions

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Abstract
Purpose Arteriovenous fistula (AVF) of the cauda equina (CE) fed by the proximal radicular artery (PRA) is very rare, and the differentiation from that of the filum terminale (FT) is important to avoid treatment-related injury to the CE when endovascular treatment is selected. The authors describe a case of AVF of the CE fed by the PRA, demonstrate the anatomical features and discuss the treatment precautions.
Methods A 69-year-old man presented with a transient weakness of lower limbs. Spinal angiography and magnetic resonance (MR) imaging revealed AVF, of which the feeding artery arose from the anterior spinal artery (ASA), forming the fistula at L2 level to be drained into the longitudinal venous trunk. Under a tentative diagnosis of AVF of the FT, endovascular treatment was attempted but failed due to impossible catheterization into the ASA. Therefore, surgery was performed.
Results Intraoperative finding revealed that the feeding artery and draining vein were not on the FT but on the CE, resulting in the proper diagnosis of AVF of the CE. Surgical clips were applied to the draining vein closest to the fistula, and postoperatively the symptom improved gradually. Although we thoroughly reevaluated spinal angiography and MR images postoperatively, AVF of the CE fed by the PRA and that of the FT were not distinguishable.
Conclusions The authors described a case of AVF of the CE fed by the PRA and demonstrated the difficulty of the differentiation from that of the FT. The utmost precautions are necessary when endovascular treatment is selected.

Keywords Arteriovenous fistula · Cauda equina · Filum terminale · Anatomy · Differentiation

Abbreviations

AVF	Arteriovenous fistula
ASA	Anterior spinal artery
CE	Cauda equina
FT	Filum terminale
MR	Magnetic resonance
PRA	Proximal radicular artery
PSA	Posterior spinal artery

Introduction

Arteriovenous fistula (AVF) of the cauda equina (CE) and that of the filum terminale (FT), both of which are intradural AVFs below conus medullaris, are rare. Especially, as concerns AVF of the CE, only ten cases have been reported, and barely two cases of them were fed by the proximal radicular artery (PRA) [1–3]. We think the differentiation between AVF of the CE fed by the PRA and that of the FT is important because an unthoughtful selection of endovascular treatment can cause damage to

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the CE. However, little is discussed about the importance of the differentiation of the two entities. Here, we present a case of AVF of the CE fed by the PRA, demonstrate the anatomical features, and discuss the treatment precautions focusing on the differentiation from that of the FT.

Case report

A 69-year-old man presented with a sudden onset of 2-time transient weakness of lower limbs for the last 1 month. The patient had traumatic compression fracture of thoracic vertebra T11 1 year before.

On admission, the patient could walk by himself, but mild disability of the left knee extension, loss of pain and temperature sensation of the left anterior thigh, and frequent micturition were noticed. Sagittal T2-weighted spine magnetic resonance (MR) images revealed a high signal intense lesion in the T6 cord to the conus medullaris as a sign of chronic venous congestion, and flow voids suggesting the tortuous and dilated perimedullary vessels from T6 to L2 in addition to old compression fracture at T11 (Fig. 1).

Left T9 intercostal arteriogram demonstrated the artery of Adamkiewicz and the anterior spinal artery (ASA), and

Fig. 1 **a**, **b** Sagittal T2-weighted magnetic resonance images at admission showing a high signal intense lesion in the cord from T6 to the conus medullaris, perimedullary flow voids from T6 to L2 and old compression fracture at T11 (*white arrow*)

the ASA gave rise to the feeding artery, which we regarded as the artery of the FT, beyond the level of the conus medullaris (Figs. 2, 3). The feeding artery connected to the fistula at L2 level, and the enlarged tortuous draining vein ran upward and connected to the anterior longitudinal venous trunk (Figs. 2, 3). Other intercostal, lumbar and sacral arteriograms showed no abnormalities. Under a tentative diagnosis of AVF of the FT, as we speculated that the feeding artery was the artery of the FT and that the AVF existed on the FT, endovascular obliteration of the fistula was attempted under local anesthesia, preparing for provocation tests. However, catheterization into the ASA was failed, and therefore direct surgery was performed.

Under intra-operative monitoring of motor evoked potentials and electromyogram of the external anal sphincter, a total laminectomy of L2 and a partial laminectomy of L3 were performed. After opening the dura mater, the FT was easily identified, but enlarged vessels were not found on it (Fig. 4a). Advancing through the CE revealed the enlarged draining vein on a CE nerve root (Fig. 4b). With indocyanine green videoangiography and Doppler ultrasonography, we identified the feeding artery, which was not the artery of the FT but the PRA, the fistula and the draining vein (Fig. 4c). Because the vessels and nerve roots adhered to each other around the fistula, the draining vein was clipped as near the fistula as possible.

The postoperative course was uneventful and the symptoms improved gradually. On postoperative spinal angiography, the feeding artery, the PRA, was invisible at the bottom of the conus medullaris and the draining vein was not demonstrated (Fig. 5).

Discussion

The angioarchitecture of the CE and the FT are not familiar because of the rarity of their vascular lesions and radiologically difficult visualization [4]. Although the CE and the FT lie side by side, their angioarchitectures are different from each other because of the embryological reasons. During development, the caudal end of the spinal cord dedifferentiates and forms the ependymal tube covered with a thin layer of marginal or fibrous layer, which is the FT; and therefore, embryologically, the FT is a part of the spinal cord [5]. On the other hand, the CE is the collective bundle of dorsal and ventral lumbosacral spinal nerve roots that have been drawn inferiorly during development, and it belongs to peripheral nervous system [4, 6].

The artery of the FT arises from the bifurcation of the ASA which constitute the arterial basket at the conus medullaris [5]. It descends on the FT, and diminishes considerably at its first few centimeters, where the artery branches arterioles which supply the coccygeal nerve roots

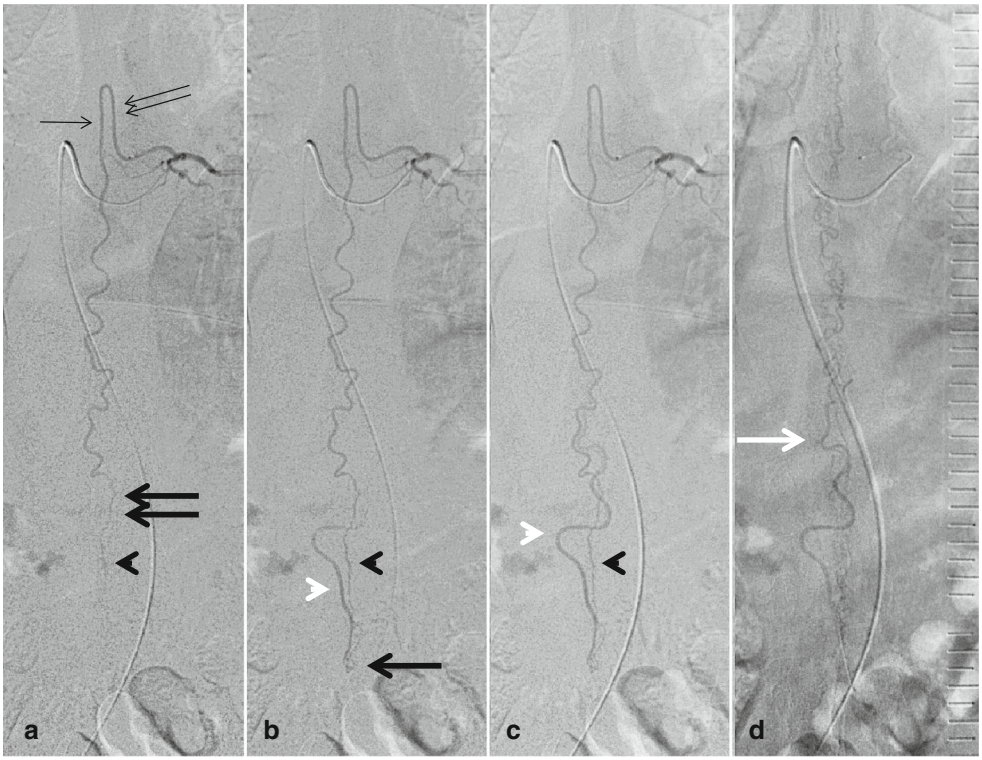

Fig. 2 **a–d** Left T9 intercostal angiogram in anteroposterior view showing the artery of Adamkiewicz (*black thin double arrow*), the anterior spinal artery (ASA) (*black thin arrow*), the feeding artery (*black arrow head*) arising from the ASA beyond the level of conus medullaris (*black thick double arrow*), arteriovenous fistula (*black thick arrow*), the draining vein (*white arrow head*) and the anterior venous trunk (*white thick arrow*) (**a** early phase; **b**, **c** middle phase; **d** late phase)

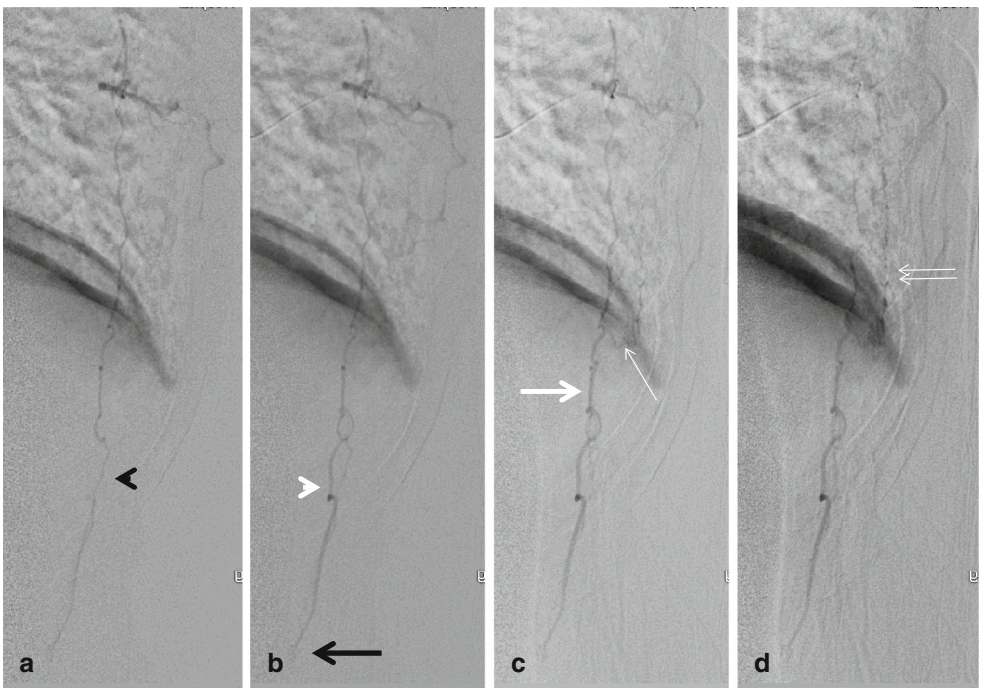

Fig. 3 **a–d** Left T9 intercostal angiogram in lateral view showing the feeding artery (*black arrow head*), arteriovenous fistula (*black thick arrow*), the draining vein (*white arrow head*), the anterior venous trunk (*white thick arrow*), venous anastomosis between the anterior and the posterior venous trunk (*white thin arrow*), and the posterior venous trunk (*white thin double arrow*) (**a** early phase; **b**, **c** middle phase; **d** late phase)

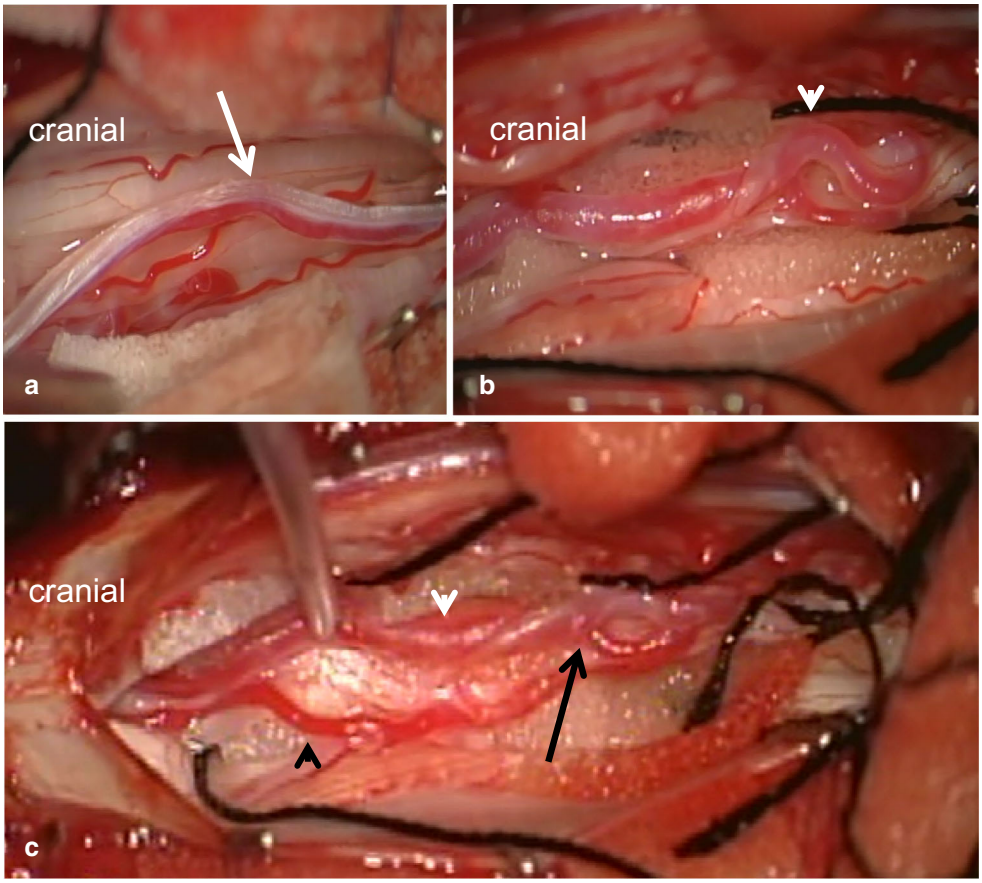

Fig. 4 Intraoperative findings. **a** Enlarged vessels are not found on the filum terminale (*white arrow*); **b** the enlarged draining vein (*white arrow head*) is observed on a cauda equina (CE) nerve root; **c** the feeding artery (*black arrow head*), the arteriovenous fistula (*black arrow*) and the draining vein are shown on a CE nerve root. *Cranial* cranial side

adherent to the FT [4, 5]. The vein of the FT exists constantly on the ventral aspect of the FT, behind the artery of the FT, and is continuous from the anterior longitudinal venous trunk to the sacral dural sac, traversing the dura to join the sacral venous plexus [4, 5].

On the other hand, angioarchitecture of the CE is more complicated. The radiculomedullary and radiculopial arteries course along each CE nerve roots and join the ASA, the posterior spinal artery (PSA) or the vasa corona; however, they do not give any branches to the CE nerve roots in their mid-course [4, 6]. The ASA, the PSA or the vasa corona give rise to the PRAs in the proximal CE nerve roots, and the segmental arteries give rise to the distal radicular arteries in the distal CE nerve roots [4, 6]. The CE nerve roots are accompanied by segmentally arranged veins in varying size that are derived from either the anterior or posterior longitudinal venous trunk, although the venous anatomy of the CE is still unknown [4].

In our case, the feeding artery was demonstrated to run continuously and downwardly from the ASA beyond the level of the conus medullaris; however, because we could not identify the arterial basket even with thorough evaluation of spinal angiography, it was not distinguishable whether the feeding artery was branched from the arterial basket, the ASA before the arterial basket or the vasa corona. When the feeding artery is branched from the arterial basket, it is more likely to be the artery of the FT. On the other hand, when the feeding artery is branched from the ASA before the arterial basket or the vasa corona, it is more likely to be the PRA, meaning that AVF exists on the CE. The draining vein ran upward and connected to the anterior venous trunk in our case. As both veins of the FT and venous system of the CE are continuous from anterior venous trunk, differentiation by venous anatomy is difficult [4, 5].

In general terms, we think that the accurate differentiation between AVF of the CE fed by the PRA and that of the FT is difficult, even with thorough evaluation of spinal angiography. Chanthanaphak et al. [7] described that demonstration of enlarged ASA and artery of the FT suggested AVF of the FT; however, they did not mention AVF of the CE, and there is no guarantee that the enlarged feeding artery below conus medullaris is not the PRA, but the artery of FT. Hong et al. [3] reported seven cases of

Fig. 5 Three-dimensional rotational angiography at admission (**a**) and after surgery (**b**) showing that the feeding artery, the proximal radicular artery, is invisible at the bottom of the conus medullaris and that the draining vein is not demonstrated after surgery

AVF of the CE; however, they were all fed by the distal radicular artery from the lateral sacral artery of the iliac artery, and therefore the differentiation could not be matter.

For clinical features, in AVF of the CE fed by the PRA of our case and previously reported two cases, clinical symptoms were weakness and paresthesia in lower limbs and bladder dysfunction which were derived from myelopathy caused by venous congestion of the spinal cord [1]. All cases were male and the mean age was 57.0 years. Treatment was direct surgery and the symptoms improved postoperatively in all cases. Whereas, for AVF of the FT, clinical symptoms in previously reported 42 cases were weakness, paresthesia, numbness and pain in lower limbs, back pain, bowel/bladder dysfunction and sexual disturbance [3, 7–22]. Most of the cases were male (76%) and the mean age was 57.0 years. Twenty-seven of the 42 cases (64%) were treated by direct surgery, 5 (12%) by combined endovascular treatment and surgery, and 10 (24%) by endovascular treatment. Although most of the symptoms improved, bowel/bladder dysfunction and sexual disturbance tended to be difficult to improve [8, 13, 22].

As described above, direct surgery, endovascular treatment and combined endovascular treatment and surgery are reported for the treatment of AVF of the CE and that of the FT. We think that the differentiation between AVF of the CE fed by the PRA and that of the FT is very important, especially in the case of endovascular treatment, although we do not have accurate methods of differentiation. For the endovascular treatment, a microcatheter should be advanced ideally by the fistula, but it is sometimes technically difficult because of small caliber of the ASA and the long distance between the origin of the radiculomedullary artery and the fistula [8]. As a result, if glue is applied and obstructs the PRA of some length, radiculopathy, namely CE syndrome, may occur in the case of AVF of the CE [23]; in contrast, occlusion of the artery of the FT neither causes significant disabilities nor impairs patient's quality of life, although the coccygeal nerve roots may be disturbed, causing sensory disturbance around the coccyx [5]. Therefore, if the endovascular treatment is selected for the case which is difficult to differentiate between AVF of the CE fed by the PRA and that of the FT, the utmost precautions, for example carrying out provocation tests, are necessary.

Conclusion

We presented a case of AVF of the CE fed by the PRA, demonstrated the anatomical features compared to that of the FT, and discussed the difficulty of the differentiation of the two entities. Utmost precautions are necessary for endovascular treatment for such a case.

Compliance with ethical standards

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

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