

Filum Terminale Arteriovenous Fistula with Additional Arterial Supply by the Posterior Spinal Artery: A Report of Two Cases

Abstract

Filum terminale arteriovenous fistulas (FTAVFs) are rare and usually supplied by the artery of the filum terminale, distal termination of the anterior spinal artery (ASA). These fistulas may also supply from the lateral sacral artery (LSA) arising from the internal iliac artery and/or middle sacral artery. Additional supply by the posterior spinal artery (PSA) has never been mentioned before. The authors describe two middle-aged men harboring FTAVFs manifested with progressive myelopathy and bowel/bladder dysfunction. The first fistula was supplied by the ASA with additional supply by the dilated PSA connecting with the ASA. Another fistula was supplied by the LSA and the PSA communicating with the ASA. The first case was successfully treated by endovascular treatment with N-butyl cyanoacrylate through the enlarged PSA, whereas the second case was surgically treated by direct obliteration of the fistula. Both patients had good neurological outcome.

Keywords: Anterior spinal artery, filum terminale arteriovenous fistula, posterior spinal artery, the arterial basket of the conus medullaris

Introduction

Spinal cord arteriovenous malformations have been classified into four subtypes including Type I, spinal dural arteriovenous fistulas (AVFs); Type II, intramedullary glomus malformations; Type III, extensive juvenile malformations; and Type IV, intradural perimedullary AVFs. Type IV spinal cord arteriovenous malformations have been further divided into three subtypes including Type IVa, small or low-flow AVF supplied by single arterial branch of the anterior spinal artery (ASA); Type IVb, intermediated-sized fistula supplied by multiple arterial feeders; and Type IVc, giant high-flow fistula fed by several feeding vessels of the ASA and posterior spinal artery (PSA).^[1,2]

Filum terminale AVFs (FTAVFs), located below the conus medullaris along the course of the filum, are characterized by a single direct communication between the artery of the filum terminale, distal termination of the ASA, and the vein of the filum terminale without intervening nidus.^[3] FTAVFs are classified as Type IVa spinal cord malformations, usually located against the anterior aspect of the conus medullaris

or the filum terminale.^[4] FTAVFs are rare and account for approximately 3% of all spinal arteriovenous lesions.^[5,6] These fistulas often manifest with symptoms of congestive myelopathy secondary to venous hypertension and usually affect middle-aged men.^[3] FTAVFs may also supply from the lateral sacral artery (LSA) arising from the internal iliac artery and/or middle sacral artery (MSA).^[7,8] To the best of our knowledge, an additional supply by the PSA has never been mentioned previously. The authors described two cases of FTAVFs with additional supply from the PSA.

Case Report

Case 1

A 55-year-old male with a 2-year history of progressive numbness of the right lower extremity complained of weakness of the right leg for 4 months. He had no history of back pain or any injury. The patient went to the local hospital and was transferred to our institute for investigation. One month before referring, he developed urinary incontinence and constipation. The neurological examination revealed paresis of the right

**Prasert
Iampreechakul,
Naruedol Yaisarn,
Korrapak
Wangtanaphat,
Punjama
Lertbutsayanukul¹,
Somkiet
Siriwimonmas²,
Nawaporn
Techataweewan³**

Departments of Neurosurgery and ¹Neuroradiology, Prasat Neurological Institute, ²Department of Radiology, Bumrungrad International Hospital, ³Department of Anatomy, Faculty of Medicine, Khon Kaen University, Bangkok, Thailand

Address for correspondence:
Dr. Prasert Iampreechakul,
312 Rachawithi Road, Khwaeng
Thung Phaya Thai, Bangkok
10400, Thailand.
E-mail: bangruad@hotmail.com

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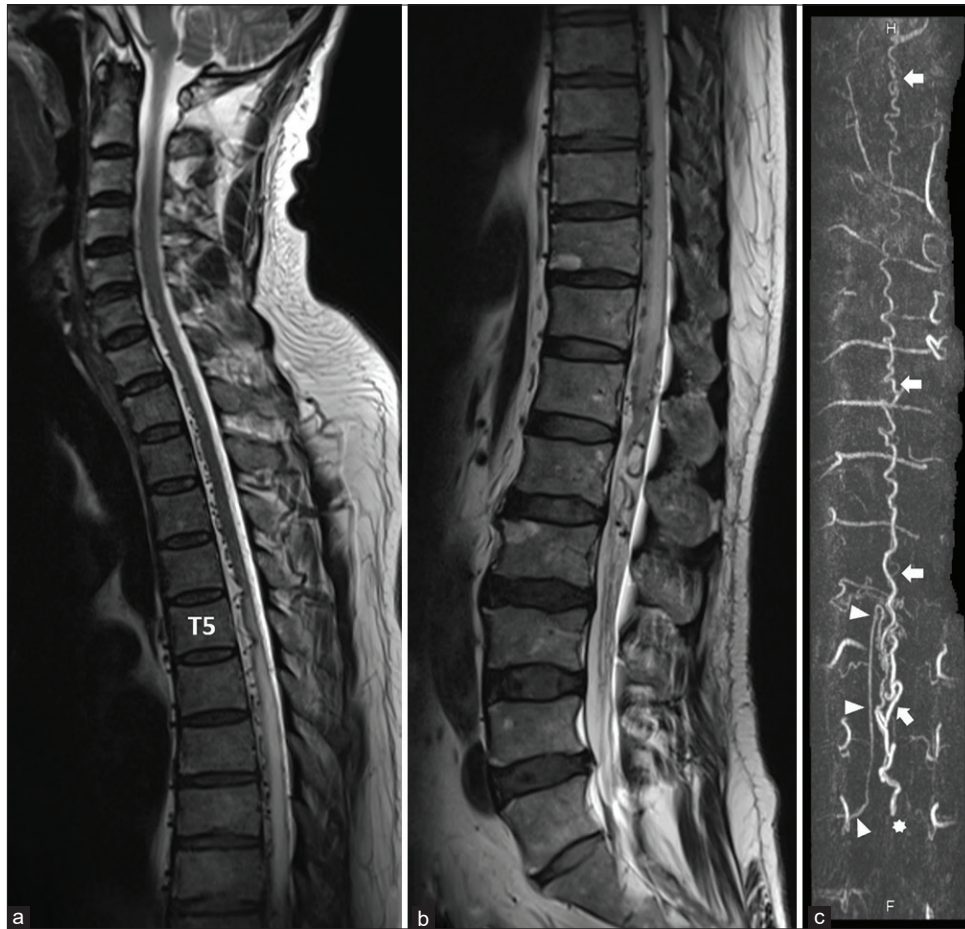


Figure 1: Sagittal T2-weighted images of the cervicothoracic (a) and thoracolumbar (b) spine reveal abnormal hyperintense T2 signal representing spinal cord congestion extending from the conus medullaris to the level of T5 and intradural flow voids along the anterior surface of the spinal cord. (c) Contrast-enhanced magnetic resonance angiography of the thoracolumbar spine demonstrates hypertrophic posterior spinal artery (arrowheads) arising from the L3 segmental artery, fistulous point (asterisk), and tortuous and enlarged intradural vessels (arrows) in the midline location extending from the level of L3 to upper thoracic level

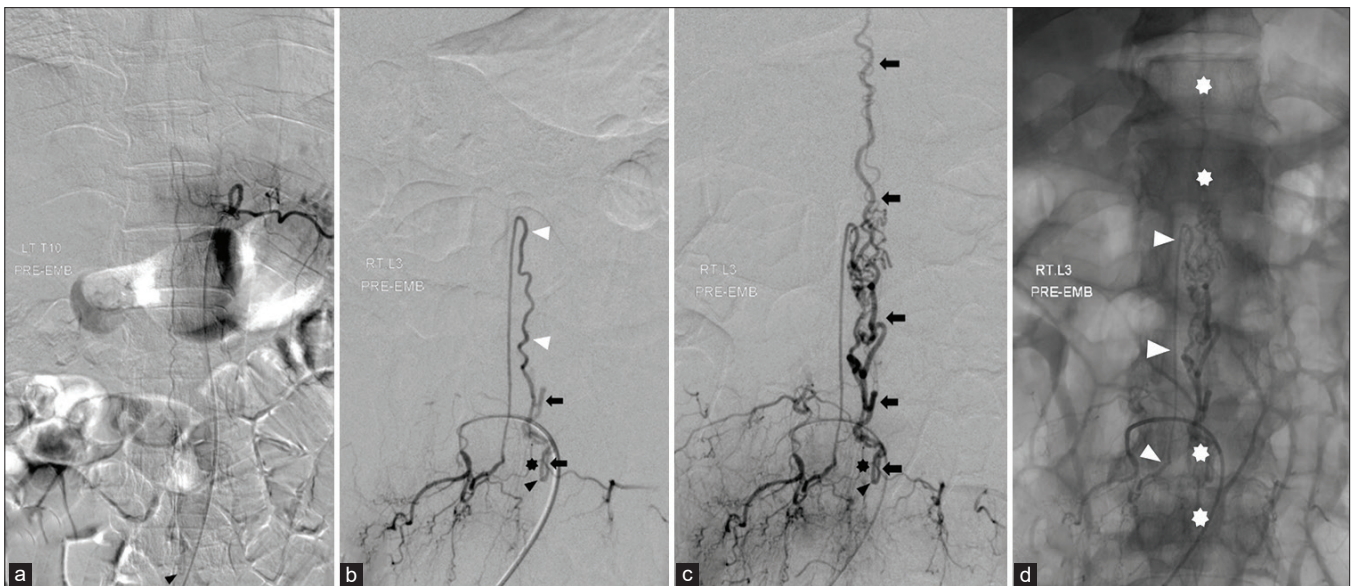


Figure 2: (a) Anteroposterior views of the left T10 intercostal artery angiography demonstrates filling of the anterior spinal artery supplying the fistula (black arrowhead) located in the filum terminale at the level of L3. Anteroposterior views of the right L3 segmental artery angiography in (b) arterial phase and venous phase (c) with and (d) without subtraction reveal the dilated posterior spinal artery (white arrowheads) connecting with the anterior spinal artery (black asterisks) supplying an arteriovenous fistula of the filum terminale (black arrowheads) with cranial drainage into the dilated vein of the filum terminale connecting to the medullary veins (arrows). The posterior spinal artery is located laterally to the spinous process (white asterisks).

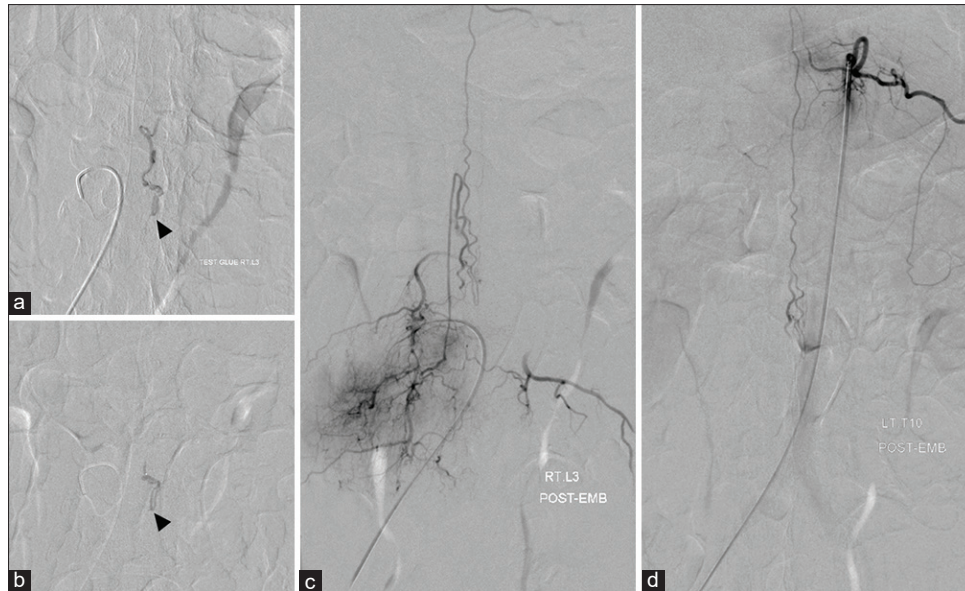


Figure 3: (a) Selective angiography with the microcatheter through the posterior spinal artery clearly demonstrates the fistulous point (arrowhead) and proximal draining vein. (b) During embolization, the glue cast can occlude the fistula and the proximal draining vein. Anteroposterior views of the right L3 (c) segmental and left T10 (d) intercostal arteries angiographies show complete obliteration of the filum terminale arteriovenous fistula with preservation of the posterior spinal artery and an anastomotic basket forming from the anterior and posterior spinal arteries

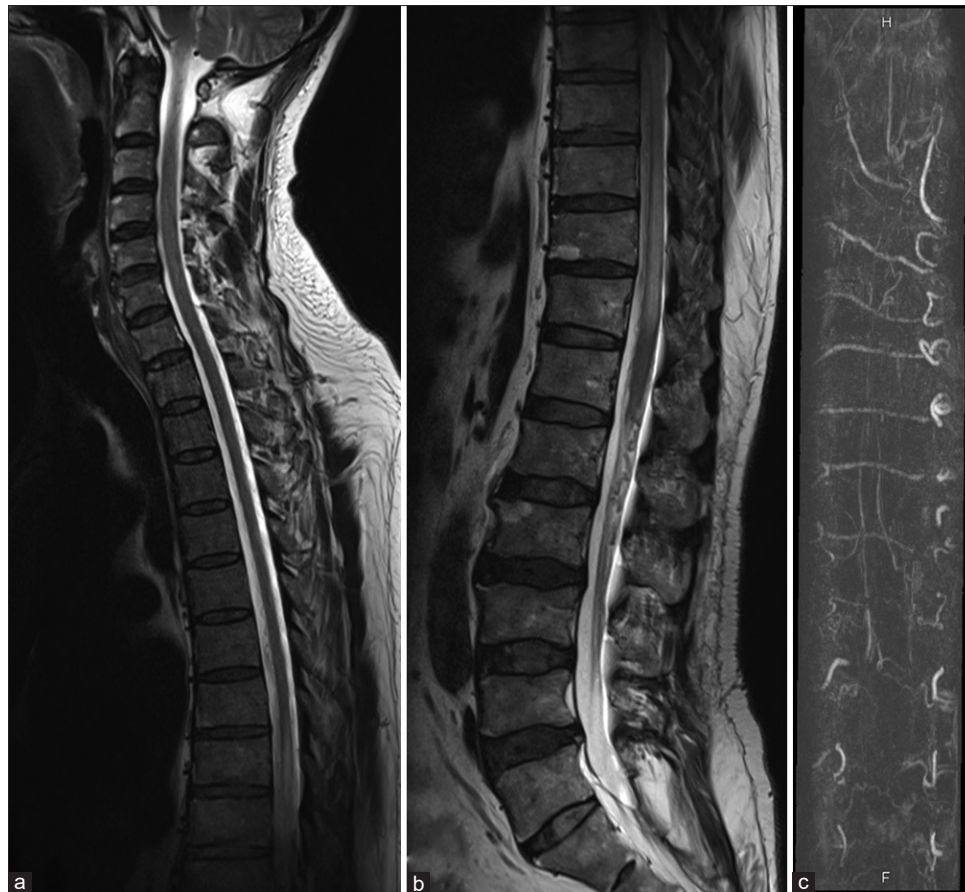


Figure 4: Sagittal T2-weighted images of the cervicothoracic (a) and thoracolumbar (b) spine, obtained 6 weeks after embolization, reveal the disappearance of intradural flow voids and significant resolution of spinal cord congestion. (c) Follow-up contrast-enhanced magnetic resonance angiography of the thoracolumbar spine confirms complete obliteration of the fistula



Figure 5: (a) Sagittal T2-weighted image of the thoracolumbar spine reveals spinal cord congestion extending from the conus medullaris to T8 level and intradural flow voids along the posterior surface of cord. There is spondylolisthesis L4-5, causing severe spinal stenosis. (b) Contrast-enhanced magnetic resonance angiography of the thoracolumbar spine demonstrates tortuous and enlarged intradural vessels (arrowheads) in the midline location extending from the level of L4-5 to mid-thoracic level

lower extremity (muscle strength 4/5). There was a decrease in pinprick sensation over the entire right lower extremity. Hyperreflexia and the presence of the Babinski sign were observed in the lower extremities. Magnetic resonance imaging (MRI) of the thoracolumbar spine revealed abnormal hyperintense T2 signal representing spinal cord congestion extending from the conus medullaris to the level of T5 and intradural flow voids along the anterior surface of the spinal cord [Figure 1a and b]. Contrast-enhanced magnetic resonance angiography (MRA) of the thoracolumbar spine demonstrated hypertrophic PSA arising from the L3 segmental artery, fistulous point, and tortuous and enlarged intradural vessels in the midline location extending from the level of L3 to upper thoracic level [Figure 1c]. Spinal angiography showed the FTAVF at the level of L3, which is supplied by the artery of the filum terminale continuing

from the ASA originating from the left T10 intercostal artery with cranial drainage through the dilated vein of the filum terminale into the tortuous perimedullary veins [Figure 2a]. In addition, the FTAVF received additional supply from the enlarged PSA arising from the right L3 segmental artery. The PSA connected with the ASA via the arterial branch along the right-sided conus medullaris [Figure 2b,c and d]. Spinal angiography of the bilateral iliac arteries showed no arterial supply to the fistula. Due to the enlarged of the PSA, we decided to proceed with endovascular as the first choice. We used Magic microcatheter 1.2 Fr (Balt, Montmorency, France). Fortunately, it was not difficult to navigate the microcatheter through the course of the PSA and the tip of microcatheter could be wedged stably the target point just proximal to the fistula. Without heparinization, transarterial embolization with N-butyl cyanoacrylate (NBCA) through the PSA was successfully performed with reaching the proximal draining vein [Figure 3a and b]. Mixture of NBCA and an oil-based contrast agent (Lipiodol Ultra Fluid; Guerbet, Aulnay-sous-Bois, France) was prepared in proportions of 1:0.7 ratio of NBCA to Lipiodol. Spinal angiography after embolization demonstrated complete obliteration of the filum terminale arteriovenous fistula with preservation of the PSA and an anastomotic basket forming from the anterior and posterior spinal arteries [Figure 3c and d]. After endovascular treatment, the patient had gradually improved. Follow-up MRI and MRA of the thoracolumbar spine, obtained 6 weeks after embolization, confirmed complete obliteration of the fistula the disappearance of intradural flow voids and significant resolution of spinal cord congestion [Figure 4]. At the 4-month follow-up, he had completely recovered without urinary incontinence and constipation.

Case 2

A 58-year-old male with a 3-year history of low back pain radiating to both legs complained of progressive weakness and numbness of lower extremities for 3 months. He had no prior history of any injury. Urine retention and constipation were noticed 1 month before the hospitalization. He was unable to walk more than about 100 meters without stopping to rest. The neurological examination revealed the evidence of spastic paraparesis (muscle strength 4/5) and the lack of pinprick sensation below L1 level. Hyperreflexia and the presence of the Babinski sign were observed in the lower extremities. MRI of the lumbosacral spine revealed abnormal hyperintense T2 signal with patchy enhancement representing spinal cord congestion extending from the conus medullaris to the level of T8. There were abnormal intradural flow voids on the posterior surface of the spinal cord from the level of T5 to the conus medullaris. Furthermore, there were herniated nucleus pulposus at the level of L2-S1, thickened ligamentum flavum with hypertrophic facet joints at the level of L3-5, and grade I degenerative spondylolisthesis

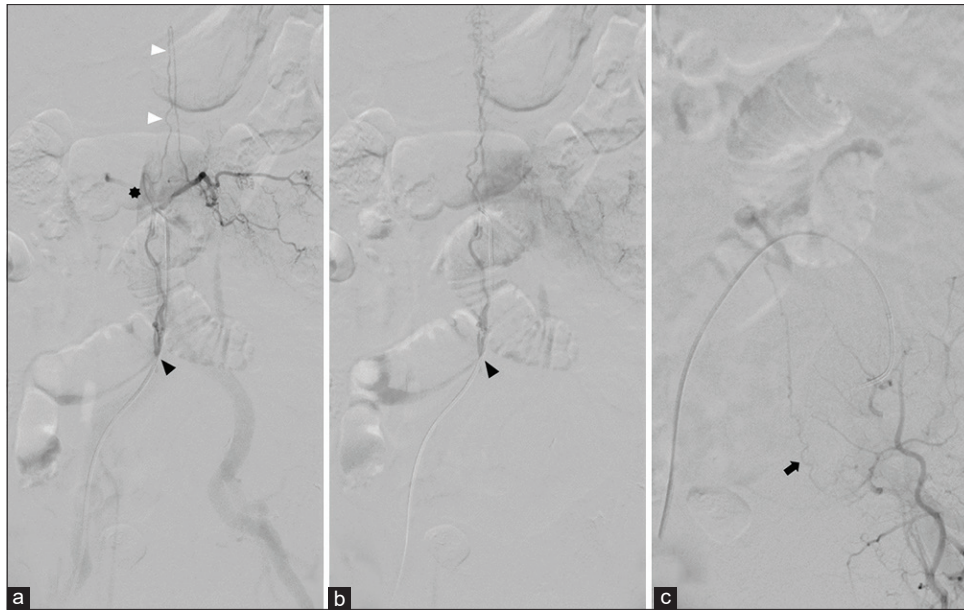


Figure 6: Anteroposterior views of the left L2 segmental artery angiography in arterial (a) and venous (b) phases show the posterior spinal artery (white arrowheads) connecting with the anterior spinal artery (asterisk) supplying an arteriovenous fistula of the filum terminale (black arrowheads) with cranial drainage into the dilated vein of the filum terminale connecting to the medullary veins. (c) Anteroposterior view of the left internal iliac artery angiography demonstrates an additional supply from a tiny branch of the left lateral sacral artery (arrow)

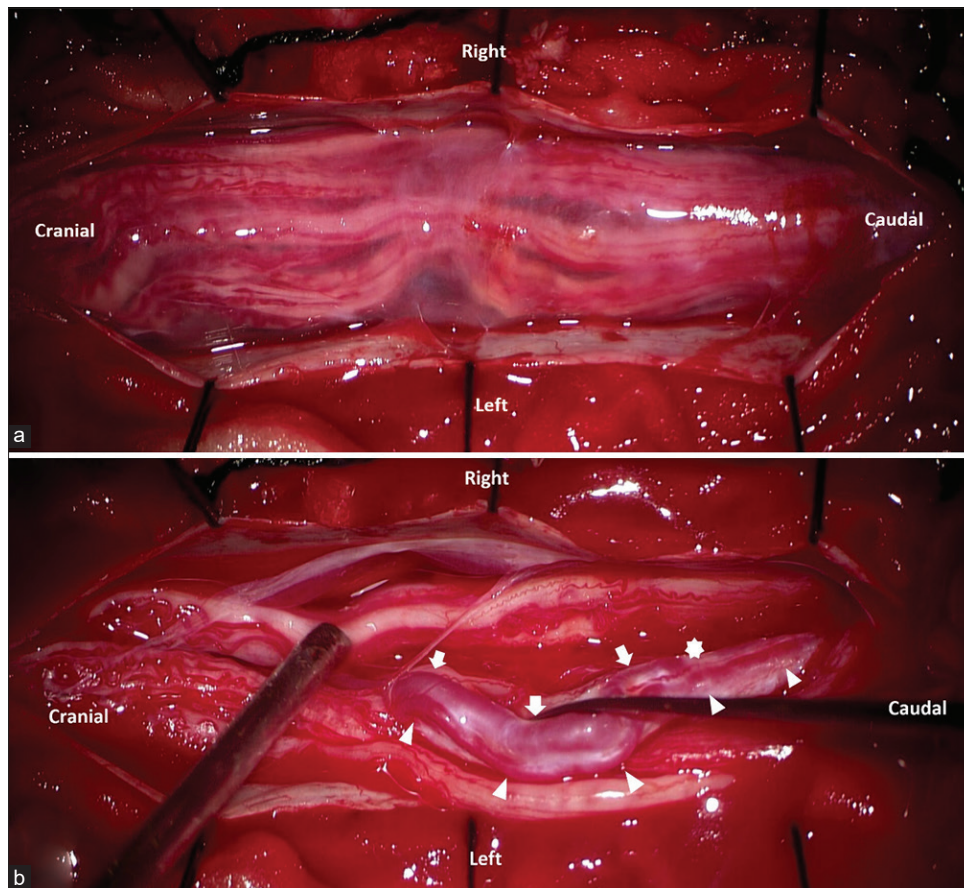


Figure 7: Intraoperative photographs during surgery on prone position. (a) After durotomy, the arachnoid membrane was found to be thickened and opaque. The roots of the cauda equina were matted together. (b) After lysis of adhesions between the nerve roots, the fistula (asterisk) was located on the thickened filum terminale, and the artery of the filum terminale (arrowheads) run parallel with the engorged vein of the filum terminale (arrows)

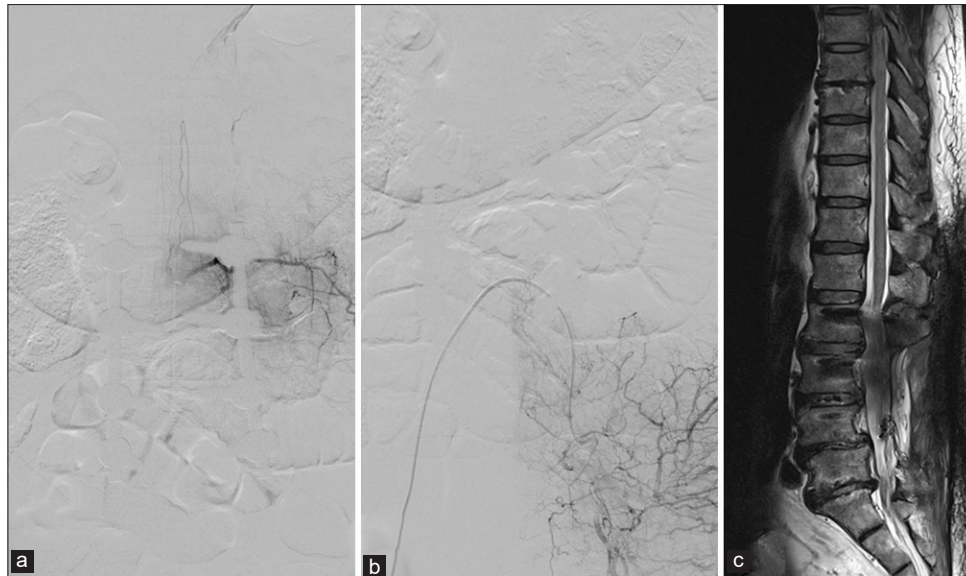


Figure 8: Spinal angiography obtained 2 weeks after the operation, anteroposterior views of the left L2 segmental (a) and left internal iliac (b) arteries injections confirm complete obliteration of an arteriovenous fistula of the filum terminale. (c) Sagittal T2-weighted image of the thoracolumbar spine obtained 3 months after the surgery demonstrates the resolution of spinal canal stenosis with disappearance of spinal cord congestion and the intradural flow voids

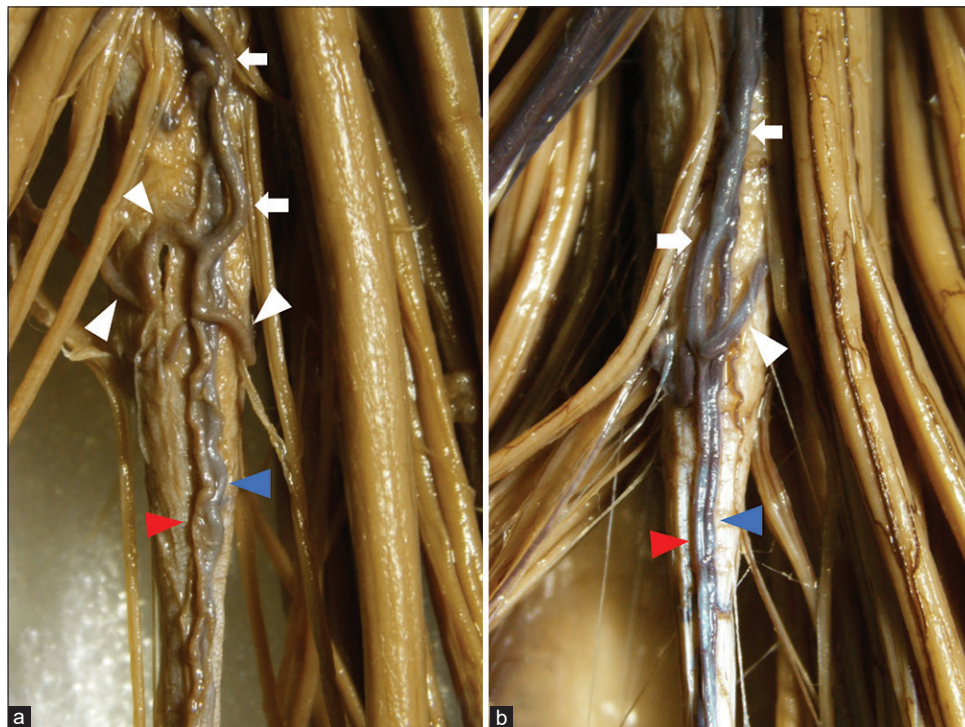


Figure 9: (a and b) Anterior gross anatomical specimens of spinal cords at the level of the conus medullaris demonstrate the connection of the anterior spinal artery (arrows) and the posterior spinal arteries (white arrowheads) forming the arterial basket of the conus medullaris giving rise to the artery of the filum terminale (red arrowheads) running parallel with the vein of the filum terminale (blue arrowheads)

at the level of L4-5 and hypertrophic facet joints, causing severe spinal canal stenosis at L4-5 compressing the cauda equina [Figure 5a]. Contrast-enhanced MRA of the thoracolumbar spine demonstrates tortuous and enlarged intradural vessels in the midline location at the level of L4-5 [Figure 5b].

Spinal angiography demonstrated the FTAVF at the level of L4-5, which is supplied by the PSA, originating from the left L2 segmental artery, connecting with the ASA giving rise the artery of the filum terminale supplying the fistula with cranial drainage into the dilated vein of the filum terminale connecting to the

medullary veins [Figure 6a and b]. The fistula also fed by the left LSA, originating from internal iliac artery [Figure 6c]. The patient underwent surgical treatment. After durotomy, the arachnoid membrane was found to be thickened and opaque. The filum terminale was adhered within the clumping of the cauda equina nerve roots [Figure 7a]. Following lysis adhesions and displacement of nerve roots, the FTAVF was identified and obliterated with small silver vessel clips [Figure 7b]. His postoperative course was uneventful. Spinal angiography obtained 2 weeks after the operation confirmed the disappearance of the fistula [Figure 8a and b]. Three months after the surgery, follow-up MRI of the lumbosacral spine also confirmed the regression of spinal cord congestion [Figure 8c]. The patient had gradually improved until being able to walk independently 6 months later.

Discussion

FTAVFs are usually classified as intradural ventral AVFs or type IVa perimedullary fistulas, located ventrally and in the midline. Intradural ventral AVFs consist of a direct fistulous connection between the anterior or PSA and a pial vein.^[1,9] Type IVa perimedullary fistulas are typically slow-flow lesions and usually located on the ventral surface of the conus medullaris or filum terminale. The arterial supply to the perimedullary fistula of the conus medullaris arise from the ASA and/or PSA, whereas the fistula of the filum terminale usually come from the ASA.^[10] According to a series of type IV perimedullary fistulas studied by Cho *et al.*,^[11] this type of the fistulas was found in younger age group with mean age 28 years. Whereas FTAVFs usually encountered in middle-age patients.^[3,12] In addition, type IVa spinal cord arteriovenous malformations may present with subarachnoid hemorrhage and/or hematomyelia.^[2] On the contrary, hemorrhagic events have never been reported from FTAVFs.^[3,8,13] Therefore, we have believed that FTAVF should be categorized separately from type IV spinal arteriovenous malformations.

Based on the study of the vascularization of the filum terminale by Djindjian *et al.*,^[7] the artery of the filum terminale derives from the termination of the ASA and traveled initially on the ventral aspect of the conus medullaris before descending on the filum with decreasing progressively in diameter. In addition, the proximal portion of the artery of the filum terminale gives origin in arterioles supplying the coccygeal nerve roots adherent to the filum. The filum terminale may also supply from the LSA arising from the internal iliac artery and/or MSA. The vein of the filum terminale, always larger than the artery of the filum terminale, is located on the ventral aspect of the filum behind the artery of the filum terminale throughout its course on the filum. The bidirectional venous drainage of this vein includes descending toward the sacral venous plexuses and ascending toward the perimedullary veins through

anterior and posterior spinal veins or the juxtamedullary anastomoses at the level of the conus medullaris. No vessels were found on the dorsal surface of the filum terminale.

At the level of the conus medullaris, the ASA may form an anastomotic basket with the PSAs via anastomotic branches. The arterial basket of the conus medullaris consists of 1 (unilateral) or 2 (bilateral) arterial branches circumferentially connecting the ASA and PSAs.^[14] We collaborated with department of anatomy from Khon Kaen University for study of the ASA in 100 Thai human cadaveric spinal cords (unpublished data). The circumferential anastomosis of the ASA and PSAs through the arterial branches of arterial basket around the conus medullaris were found in many specimens [Figure 9]. However, FTAVF with an additional supply by the PSA has never been reported previously.^[3,8,12,13,15-17] In the present study, we primarily demonstrated 2 patients harboring FTAVFs with additional supply from the PSAs communicating with the ASAs via the arterial branches of the arterial basket of the conus medullaris.

FTAVFs can be treated by surgery, endovascular treatment, or both. The goal of treatment is complete obliteration of the fistula with preservation of normal arterial supply to spinal cord.^[2] The key to complete occlusion is obliteration of the proximal vein.^[3] Even through surgical procedure is more invasive than endovascular treatment, surgical treatment has been the preferred method of treatment with higher complete obliteration rates.^[5] Furthermore, surgical obliteration of FTAVFs is technically simple and highly effective with low rate of recurrence.^[4,12,13] Endovascular treatment should be considered as second-line choice because of the difficulty in navigating a microcatheter through the long and tortuous course of the thin ASA; the possibility of reflux of the liquid embolic material into the ASA; the risk of tearing, dissecting, thrombosis, or vasospasm of the ASA during embolization; and requiring expertise and experience in neurointerventional procedure. In addition, endovascular has a chance of recanalization or recurrence of the fistula more than surgery.^[2,3,5,18] In the present study, we decided to try endovascular treatment as first choice in the first case because there was the accessible dilated PSA connecting with the artery of the filum, the termination of the ASA, through the arterial basket of the conus medullaris. The important factor for the successful transarterial embolization is an introduction of the tip of microcatheter in a more stable and distal position to the shunt point. During embolization with NBCA, the safety margin for glue reflux was short. The glue should be penetrated the artery of the filum close to the fistula without reflux into the ASA. In addition, the glue should be stopped just the proximal draining vein for avoiding anterograde

venous occlusion. The heparin and steroid should be used in case of glue reflux in the ASA or anterograde venous occlusion. However, we did not use the heparin or steroid in our first case due to the successful injection of the glue penetrating just proximal vein without reflux into the ASA or PSA. In the second case, we decided to proceed with surgery due to the long course of the small PSA connecting with the thin ASA.

Conclusion

FTAVF may receive additional supply from the PSA through the arterial basket of the conus medullaris, which is an anastomotic network connecting the ASA and PSAs. Successful treatment required a precise understanding of the vascular anatomy around the conus medullaris and angioarchitecture of FTAVF.

Consent

The patients have given consent to be enrolled and have their data published.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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