



Case Report

Cauda equina arteriovenous fistula supplied by proximal radicular artery and concomitant sacral dural arteriovenous fistula: A case report and literature review

Prasert Iampreechakul¹, Punjama Lertbutsayanukul², Somkiet Siriwimonmas³

¹Department of Neurosurgery, Prasat Neurological Institute, ²Department of Neuroradiology, Prasat Neurological Institute, ³Department of Radiology, Bumrungrad International Hospital, Bangkok, Thailand.

E-mail: *Prasert Iampreechakul - bangruad@hotmail.com; Punjama Lertbutsayanukul - lpunjama@hotmail.com; Somkiet Siriwimonmas - somkietsiri@hotmail.com



***Corresponding author:**

Prasert Iampreechakul,
Department of Neurosurgery,
Prasat Neurological Institute
Bangkok, Thailand.

bangruad@hotmail.com

Received : 20 June 2021

Accepted : 21 July 2021

Published : 16 August 2021

DOI

10.25259/SNI_612_2021

Quick Response Code:



ABSTRACT

Background: Cauda equina arteriovenous fistulas (AVFs) fed by the proximal radicular artery are exceedingly rare. Spinal dural arteriovenous fistulas (DAVFs) in the sacral region are rare and usually misdiagnosed. We report a case of a cauda equina AVF with concomitant sacral DAVF. We also review the coexistence of multiple types of spinal vascular malformations in a single patient.

Case Description: A 54-year-old man presented with progressive weakness of the lower extremities for 1 month. Magnetic resonance imaging (MRI) of the lumbosacral and thoracic spine showed spinal cord congestion, extending from the conus medullaris to the level of T7, and abnormal tortuous and dilated flow void, running from the level of L5 to T12 along anterior surface of the spinal cord. Spinal angiography demonstrated the fistula at the level of L2 below the conus medullaris. Based on intraoperative findings, the cauda equina AVF supplied by the proximal radicular artery with cranial drainage through the enlarged radicular vein was confirmed and successfully obliterated. Another enlarged arterialized radicular vein running parallel to another cauda equina nerve root is observed with unknown origin. After the operation, the patient showed mild improvement of his symptoms. Follow-up MRI and contrast-enhanced MR angiography revealed another sacral DAVF vascularized by the lateral sacral artery.

Conclusion: The coexistence of different spinal vascular malformations in a same patient is extremely rare. Most authors of several studies hypothesized that venous hypertension and thrombosis due to the presence or treatment of the first spinal vascular lesion may produce a second DAVF.

Keywords: Cauda equina arteriovenous fistula, Filum terminale arteriovenous fistula, Multiple spinal vascular malformations, Radicular arteriovenous fistula, Sacral dural arteriovenous fistula

INTRODUCTION

Spinal vascular malformations have been classified into four subtypes including Type I, spinal dural arteriovenous fistulas (SDAVFs); Type II, intramedullary glomus malformations; Type III, extensive juvenile malformations; and Type IV, intradural perimedullary arteriovenous fistulas (PMAVFs).^[1]

Cauda equina arteriovenous fistulas (AVFs) or radicular AVFs (rAVFs) are fistulas located on a nerve root of the cauda equina intradurally and quite rare.^[5] The cauda equina nerve root

is supplied either by the distal radicular artery from the segmental artery or the proximal radicular artery from the anterior spinal artery (ASA), posterior spinal artery (PSA), or the vasa corona.^[14] Cauda equina AVFs fed by the proximal radicular arteries are extremely rare.^[13,17,22]

SDAVFs are the most common type of spinal vascular malformations.^[21] SDAVFs in the sacral region are rare and usually misdiagnosed.^[3] The incidence of sacral DAVFs is 12.5% of all SDAVFs.^[8] The standard spinal angiography may not identify sacral DAVFs unless selective injections of the internal iliac arteries are carefully examined at the time of angiography.^[3]

The coexisting of the same type of spinal vascular malformations has been reported.^[11,18] According to the large series of spinal cord arteriovenous malformations (AVMs) by Matsumaru *et al.*,^[11] they reported multifocal spinal cord AVMs in 16% and most of the cases were metameric associations. Double SDAVFs are rare and account for approximately 1–2% of all SDAVFs.^[18] The occurrence of two different types of spinal vascular malformations coexisting in the same patient is exceedingly rare.^[2,6,7,10,12,15]

The authors described a patient with concomitant cauda equina AVF fed by proximal radicular artery and sacral DAVE. The authors also reviewed the published case reports and series that enough clinical description and clearly demonstrated figures of the coexistence of multiple types of spinal vascular lesions. This literature search was performed on the Ovid MEDLINE, PubMed, Cochrane Database of Systemic Reviews, and Google Scholar with the following key words of “concomitant,” “multiple,” “coexistence,” “concurrent,” “spinal,” “vascular malformations,” and “AVF” Exclusion criteria included non-English-language articles, commentaries, and information from expert opinions.

CASE DESCRIPTION

A 54-year-old man, heavy smoker without underlying disease, was admitted to the local hospital due to progressive weakness of the lower extremities for 1 month. He had no history any injury. Three months earlier, he experienced low back pain radiating to both legs, predominantly affecting the left side. Furthermore, difficulty in urination and constipation were observed 1 week before admission. Magnetic resonance imaging (MRI) of the lumbosacral and thoracic spine revealed abnormal hyperintense T2 signal, representing spinal cord congestion, extending from the conus medullaris to the level of T7. There was abnormal tortuous and dilated flow void, running from the level of L5 to T12 along anterior surface of the spinal cord [Figure 1]. A preliminary diagnosis was SDAVF. The patient was transferred to our institute and admitted for further investigation. The neurological examination revealed the evidence of spastic paraparesis



Figure 1: Sagittal T2-weighted image of the thoracolumbar spine shows abnormal hyperintense T2 signal representing spinal cord congestion extending from the conus medullaris to the level of T7 and intradural flow void from the level of L5 to T12 along anterior surface of the lower spinal cord.

(muscle strength 4/5), the lack of pinprick sensation below L2 level, hyperreflexia, and presence of Babinski sign in the lower extremities.

Spinal angiography demonstrated the fistula at the level of L2 below the conus medullaris, which is supplied by the PSA originating from the left L1 segmental artery with cranial drainage through the paralleling dilated vein into perimedullary vein. Without selective catheterization into both internal iliac arteries, lower aorta and bilateral common iliac arteries angiography reveals no more supply to the fistula [Figure 2]. The ASA arose from the left T6 intercostal artery without supplying to the fistula. Initially, we interpreted that this fistula was filum terminale AVF (FTAVF) which is fed by the ASA supplying from the PSA via the vasa corona. Due to small and long distance of the feeder, the patient underwent surgical treatment. On prone position, total laminectomy of L2 and partial laminectomy of L3 were carried out. After durotomy, the filum terminale (FT) was identified and no fistula or abnormal vessels on it. The fistula is located on the left cauda equina nerve root supplied by the proximal radicular artery with cranial drainage through the enlarged radicular vein. Another enlarged arterIALIZED radicular vein running parallel to another cauda equina nerve root is observed with unknown origin [Figure 3]. To avoid nerve root injury by heat, the dilated proximal draining vein near the fistula on the cauda equina nerve root was clipped with small silver clips without using bipolar coagulation. Another radicular vein was left for further investigation. After the operation, the patient showed mild improvement of his symptoms. He could walk with the aid of a walker. He was discharged home 7 days later due to his requesting to do some personal issues at home.



Figure 2: Anteroposterior views of the left L1 segmental artery angiography in (a) early, (b) middle, and (c) late phases demonstrate filling of the posterior spinal artery supplying the fistula (arrow), located at the level of L2 below the arterial basket of the conus medullaris forming from the bilateral posterior spinal arteries (arrowheads), with cranial drainage into perimedullary veins. (d) Anteroposterior view of flush aortography including lower aorta and bilateral iliac arteries reveals no more supply to the fistula.

Follow-up MRI and contrast-enhanced MR angiography (MRA) of the thoracolumbar spine, obtained 3 weeks after the operation, revealed mild regression of spinal cord congestion, and remaining of intradural flow void from L5 to L2. Another SDAVF was found at left S1 neural foramen supplied by the left lateral sacral artery (LSA) originating from the left internal iliac artery with venous drainage into perimedullary veins through the dilated and tortuous radicular vein, probably corresponding with another dilated arterialized radicular vein found during the operation [Figures 4 and 5]. Comparing between preoperative the left L1 segmental artery angiography and postoperative contrast-enhanced MRA, there was the same venous drainage pattern, representing sharing the common medullary venous channel [Figure 6].

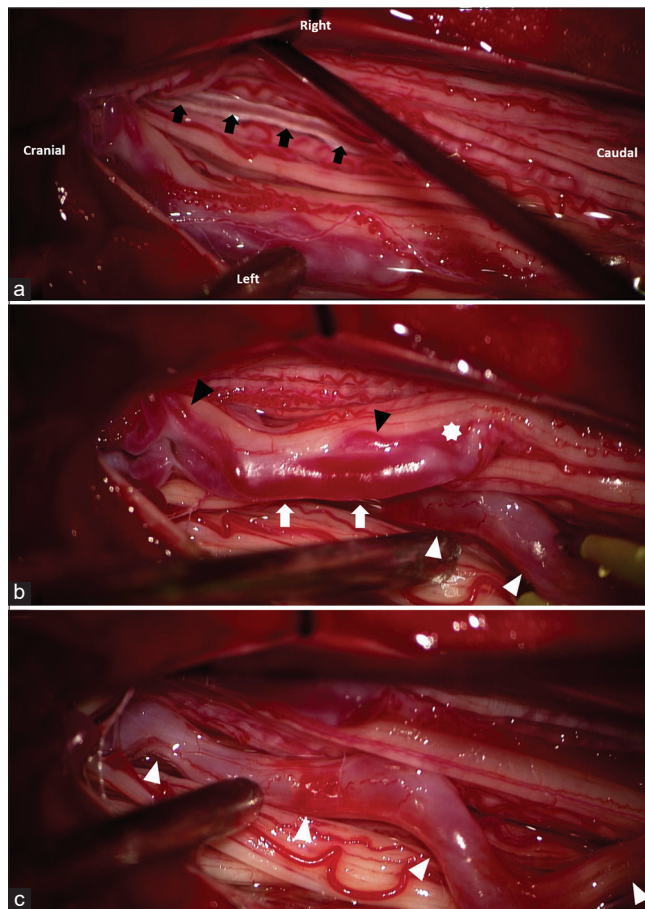


Figure 3: Intraoperative photograph during surgery on prone position after opening the dura. (a) Normal vessels on the filum terminale (black arrows). (b) The fistula is located on the left cauda equina nerve root (asterisk) supplied by the feeding artery (black arrowhead) with cranial drainage through the dilated radicular vein (white arrow) (c) another enlarged arterialized radicular vein (white arrowhead) running parallel to the cauda equina nerve root is observed with unknown origin.

Spinal angiography and probable embolization in the same setting were scheduled for another week. Few days before hospitalization for further treatment, the patient developed loss of consciousness at home and was sent to the emergency department of the local hospital and intubated promptly. Few minutes later, the patient had a cardiac arrest. Immediate cardiopulmonary resuscitation was performed unsuccessfully. Without an autopsy, the cause of death was still unknown.

DISCUSSION

The cauda equina AVF

Based on the study of vascular anatomy of the cauda equina by Namba,^[14] the proximal one-third of the nerve root is vascularized by the radicular artery arising from the vasa

corona, ASA, or PSA. The distal two-third of nerve root is supplied by the radicular artery from the segmental artery. In our case, the fistula is located on the left-sided nerve root

of cauda equina fed by the proximal radicular artery arising from the PSA.

Cauda equina AVFs supplied by proximal radicular arteries are extremely rare. After review of the literature, there were only five cases, including our one case [Table 1].^[13,17,22] All patients were men with a median age of 58 years, range 40–69 years. In 2011, Ohtonari *et al.*^[17] first reported two cases in which the fistula was located on a nerve roots of the cauda equina supplied by an anterior radicular branching from the ASA confirmed by intraoperative findings. In addition, Tanioka *et al.*^[22] reported another case of AVF of the cauda equina fed by the proximal radicular artery arising from the ASA. They found that it was difficult to differentiate this fistula from FTAVF, which is supplied by the artery of the FT with cranial drainage through a closely paralleling the vein of the filum. The fistula of the cauda equina was confirmed during the operation. Recently, Namba *et al.*^[13] demonstrated another case of cauda equina AVF vascularized by proximal radicular artery from the ASA and distal radicular artery from the LSA. Using the angiographic and MRI findings, they proposed the pathognomonic findings to differentiate cauda equina AVF from FTAVF. In cauda equina AVF, the ASA revealed change in caliber and course at the conus medullaris apex, representing the transition of the ASA into a radicular artery, located off the midline. The wavy drainage pattern of the radicular vein may also be pathognomonic for cauda equina AVF. Detection by thin-section axial MRI, a draining vein joining the spinal cord above the conus medullaris apex was especially suggestive of a radicular vein.

Hong *et al.*^[5] reported seven cases of intradural arteriovenous shunts of nerve root of the cauda equina located between L4 and S1 off the midline. All radicular shunts in their study were supplied by distal radicular arteries originating from the LSAs.



Figure 4: (a) Sagittal T2-weighted image of the thoracolumbar spine obtained 3 weeks after the operation reveals mild regression of spinal cord congestion and remaining of intradural flow void from L5 to L2. (b) Coronal magnetic resonance myelography of the lumbosacral spine demonstrates abnormal flow void running from the left sacral nerve root to the left-sided conus medullaris, probably representing a dilated radicular vein.

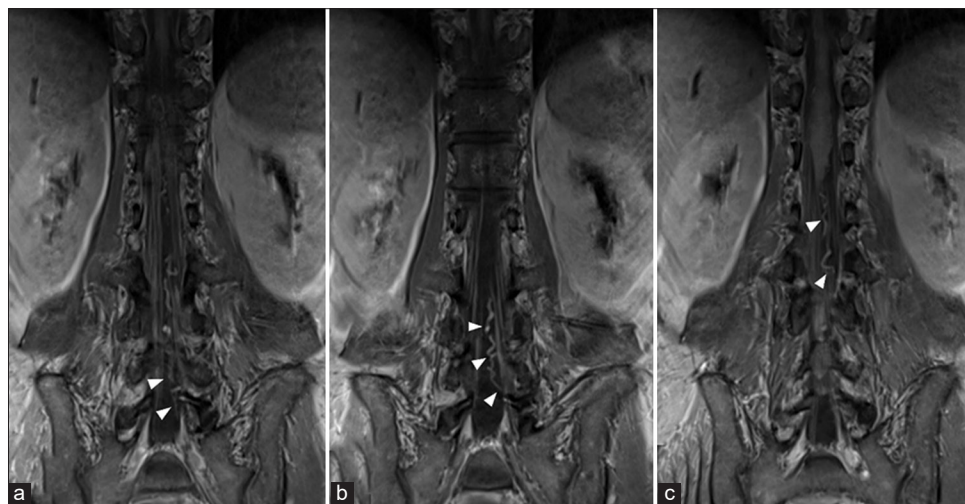


Figure 5: (a-c) Sequential contrast-enhanced T1-weighted MRI coronal images show early enhanced tortuous and dilated intradural vessel, representing a radicular vein, draining cranially from the left sacral nerve root reaching the conus medullaris.

Table 1: Literature review of patients with arteriovenous fistula of the cauda equina fed by the proximal radicular artery.

Authors	Gender/Age	Symptoms and signs	Location	Main feeder	Treatment	Neurological outcome
Ohtonari <i>et al.</i> (2011) ^[17]	M/40	Sudden weakness of his leg with paresthesia	L1-L2	ASA/Lt. T8	Surgery	GR
	M/62	Left leg hypesthesia, paraparesis dominantly in left leg, and urinary hesitation	L2	ASA/Lt. T8	Surgery	GR
Tanioka <i>et al.</i> (2018) ^[22]	M/69	Sudden onset of 2-time transient weakness of lower limbs for the last 1 months	L2	ASA/Lt. T9	Surgery	GR
Namba <i>et al.</i> (2020) ^[13]	M/58	Paraparesis, sensory disturbance, dysuria	L3	- ASA via PSA to vasa corona/ Lt. T11 - Lt. LSA	Embolization (Residual fistula)	No clinical sequelae declined
Present case	M/54	A 3-month history of low back pain radiating to both legs, progressive weakness of lower extremities for 1 month, BBD	L2	PSA/Lt. L1	Surgery	PR

ASA: Anterior spinal artery, BBD: Bowel and bladder dysfunction, GR: Good recovery, L: Lumbar, LSA: Lateral sacral artery, M: Male, PR: Poor result, PSA: Posterior spinal artery



Figure 6: Comparing between anteroposterior view of (a) preoperative the left L1 segmental artery angiography in late venous phase and (b) postoperative contrast-enhanced magnetic resonance angiography demonstrates the same venous drainage pattern (arrowheads), representing sharing the common medullary venous channel. The feeder from the left lateral sacral artery (arrows) is noted.

To avoid treatment-related injury to the cauda equina during endovascular treatment, surgery is the best choice for the fistula of the cauda equina nerve root.^[13,15] In the present study, the cauda equina AVF fed by the proximal radicular artery originating from the PSA was confirmed by the intraoperative findings and was successfully obliterated by surgery.

The concomitant different types of spinal vascular malformations

Owing to the rarity of concurrent different spinal vascular malformations in a single patient, the diagnosis of two lesions at the same time remains challenging, as shown in our case. According to our review about the coexistence of multiple types of spinal vascular lesions, the collected data in this review included demographic data (i.e., patient gender and age), symptoms and signs, type with location of each fistula, synchronous or metachronous occurrence of these fistulas, the presence of sharing the same venous drainage, treatment, and neurological outcome of the patients [Table 2].^[2,4-7,9,10,12,13,15,16,19,20] Our search found 18 cases including our case with 36 spinal vascular lesions. There were 13 men (72.2%) and 5 women (27.8%). Their median age was 34.5 years (range 14–65 years). Most patients presented with back pain, sciatic pain, progressive myelopathy, and/or bowel/bladder dysfunction. Five patients presented with hemorrhagic event. Two patients were associated with congenital anomaly. Of 36 spinal vascular lesions, 13 lesion (36.1%) were AVM or PMAVF at or around the conus medullaris, 9 (25%) SDAVFs at the lumbar or sacral region, 4 (11.1%) SDAVFs at thoracic region, 4 (11.1%) cauda equina or radicular AVFs, 3 (8.3%) PMAVFs at the thoracic region, 2 (5.6%) FTAVFs at sacral region, and 1 (2.8%) spinal epidural AVF at lumbosacral region. The concurrent conus medullaris lesions and other spinal vascular lesions are the most common concurrent types and were found in 13 patients (72.2%). The coexistence of rAVF and other spinal vascular malformations was found in 4 patients (22.2%), including our 1 case. The occurrence of fistulas in the same patient was synchronous in 13 (72.2%), and metachronous in 5 (27.8%). In metachronous group, most patients presented

Table 2: Literature review of patients harboring the coexistence of multiple types of spinal vascular lesions.

Authors	Gender/ Age	Symptoms and signs	First lesion	Second lesion	Occurrence of the fistula	Share the same venous drainage	Treatment	Neurological outcome
Dam-Hieu <i>et al.</i> (2001) ^[2]	M/49	Back pain with bilateral sciatic pain over a 6-month period, rapidly progressive myelopathy for 2 weeks, BBD	SDAVF (T6 level)	Conus AVF	Synchronous	N/A	ST both lesions	GR
Hsu <i>et al.</i> (2002) ^[6]	M/44	Progressive weakness of both legs and urinary incontinence.	Conus AVM	Sacral DAVF of FT	Synchronous	N/A	ST both lesions	GR
Morgalla <i>et al.</i> (2003) ^[12]	M/61	A 6-month history of progressive weakness of both legs, recurrent symptoms 2 months after first surgery	SDAVF (T6 level)	PMAVF (T7 level)	Metachronous (2 months)	Yes	ST both lesions	GR
Nishio <i>et al.</i> (2003) ^[15]	F/16	Progressive back pain, weakness, and numbness of the left lower limb	Conus AVM	rAVF (L4 level)	Synchronous	N/A	Conus AVM-EVT and ST rAVF-None	N/A
Ling <i>et al.</i> (2005) ^[10]	M/14	Bilateral lower limb paraparesis, Lower back pain worsening over a 5-day period, SAH, hematomyelia, BBD in patient with Hereditary hemorrhagic telangiectasia	PMAVF (T11 level)	SDAVF (T12 level)	Metachronous (9 months)	N/A	PMAVF-ST SDAVF-EVT	GR
Krings <i>et al.</i> (2006) ^[7]	M/35	A 6-month history of progressive myelopathy	PMAVF (T5 level)	SDAVF (T6 level)	Synchronous	N/A	ST both lesions	GR
Sasaki <i>et al.</i> (2012) ^[20]	M/32	Slowly progressive paraparesis over a 6-month, recurrent weakness 2 months after operation of SDAVF	Sacral DAVF	PMAVF (L1-2 level)	Synchronous	N/A	ST both lesions	IR
Hong <i>et al.</i> (2017) ^[5]	M/34	N/A	Conus AVM	rAVF (L5 level)	Synchronous	Yes	Conus AVM-N/A rAVF-EVT	N/A
Li <i>et al.</i> (2017) ^[9]	M/65	A 4-year history of progressive weakness and numbness of both legs, BBD	SDAVF (L2 level)	FTAVF (S3 and S5)	Synchronous	Yes	SDAVF-EVT FTAVF-ST	GR

(Contd...)

Table 2: (Continued).

Authors	Gender/ Age	Symptoms and signs	First lesion	Second lesion	Occurrence of the fistula	Share the same venous drainage	Treatment	Neurological outcome
Rosi <i>et al.</i> (2018) ^[19]	M/27	Severe progressive left fluctuating lumbosciatic pain	Conus AVF	Sacral DAVF	Synchronous	Yes	Conus AVF-EVT Sacral DAVF- None*	GR
	M/16	SAH in patient with Klippel- Trenaunay syndrome	Conus AVM	Sacral DAVF	Metachronous (4 years)	Yes	EVT both lesions.	IR
	M/18	SAH	Conus AVM	Sacral DAVF	Synchronous	Yes	EVT both lesions.	GR
	F/37	Recurrent hematomyelia during second pregnancy	Conus AVF	Sacral DAVF	Synchronous	Yes	Conus AVF-ST Sacral DAVF-EVT	N/A
	F/45	Left lumbosciatic pain and SAH at 5 years after diagnosis	AVM at thoracic cord and conus	LS DAVF	Metachronous	Yes	SDAVF- EVT AVM-EVT	GR
Niu <i>et al.</i> (2020) ^[16]	F/25	Intermittent pain of the right lower limb persisting for 1 month	Conus AVM	Sacral DAVF	Synchronous	No	ST both lesions	GR
Namba <i>et al.</i> (2020) ^[13]	M/61	Paraparesis, sensory disturbance, dysuria	PMAVF (L1)	rAVF (L4 level)	Synchronous	No	EVT both lesions	GR
Guédon <i>et al.</i> (2021) ^[4]	F/23	Progressive left foot step page, sensory deficit of the left lower limb, and lumbar pain. Recurrent symptoms 5 years after the last EVT	Conus AVM	LS SEAVF	Metachronous (5 years)	Yes	EVT both lesions	GR
Present case	M/54	A 3-month history of low back pain radiating to both legs, progressive weakness of lower extremities for 1 month, BBD	rAVF (L2 level)	Sacral DAVF	Synchronous	Yes	rAVF-ST SDAVF- None	PR

AVF: Arteriovenous fistula, AVM: Arteriovenous malformations, BBD: Bowel and bladder dysfunction, EVT: Endovascular treatment, F: Female, FTAVF: Filum terminale AVF, GR: Good recovery, IR: Incomplete recovery, L: Lumbar, LS: Lumbosacral, M: Male, N/A: Data not available, PMAVF: Perimedullary AVF, PR: Poor result, rAVF: Radicular AVF, SAH: Subarachnoid hemorrhage, SDAVF: Spinal dural AVF, SEAVF, Spinal epidural AVF, ST: Surgical treatment, *Spontaneous thrombosis

with recurrent symptoms after first treatment. The sharing of the same venous drainage of concurrent fistulas could identify in ten patients (55.6%). Surgical treatment of both concurrent lesions was performed in six patients, endovascular treatment of both

lesions in 4, combination of surgical and endovascular treatments of both lesions in 4, surgical or endovascular treatment of any one of lesions in 4. Most patients treated both lesions had good neurological outcome following the treatment.

The pathogenesis of the concurrent different spinal vascular malformations in different spinal levels

The relationship between the two different spinal vascular malformations remains unknown. Whether the concurrence is purely a coincidence or pathogenetic linked is unclear.^[7] SDAVFs, usually occurred in adulthood, are probably acquired lesions whereas intradural PMAVFs or AVMs, often occurred in younger patient, are congenital in nature. Both lesions seem to be quite different in nature. The formation of SDAVFs seems to be strongly associated with venous hypertension or thrombosis.^[4,18-20] Unusually, SDAVFs associated with other spinal vascular malformations in our review occurred in younger patients.

In 2001, Dam-Hieu *et al.*^[2] speculated that SDAVF may be the consequence of venous thrombosis of perimedullary veins, occurred from the conus medullaris shunt, probably inducing the formation of dural shunt. One year later, Hsu *et al.*^[6] reported concomitant conus AVM and sacral DAVF of the FT. They proposed that it is possible to produce a secondary SDAVF following change of venous pressure of the spinal cord caused by the spinal cord AVM. Another year later, Morgalla *et al.*^[12] reported metachronous occurrence of DAVF and PMAVF in the same patient during a period of 2 month. The clinical deterioration of the patient postoperatively indicated the existence of a possible second fistula. They suspected that the dural fistula may obscure the perimedullary fistula due to its higher shunt volume. The manifestation of a second fistula may be due to a decrease of flow in the common draining vein after the first fistula was closed. Few years later, Krings *et al.*^[7] speculated that the alteration in venous drainage caused by the congenital PMAVF might possibly promote the production of a second DAVF due to elevated pressure with concomitant venous stagnation and subsequent thrombosis. A decade later, Li *et al.*^[9] reported concomitant SDAVF at L2 level and FTAVF at sacral region. They hypothesized that the venous hypertension caused by the primary lesion promotes the pathologic connection the draining vein, inducing the subsequent development of the secondary lesion. Similarly, Sasaki *et al.*^[20] speculated that a single SDAVF or PMAVF is postulated to aggravate elevated pressure, venous stagnation, and subsequent thrombosis, resulting in formation of another SDAVF. In addition, Rosi *et al.*^[19] reported five cases of concomitant conus AVM or AVF and sacral DAVF and assumed that the concurrent lesions are the pathophysiological linked. Most dural fistulas were found in the area in which the venous drainage of the conus lesions ended. They proposed that the venous hypertension created by the venous drainage toward the sacral area responsible for angiogenesis inducing the dural shunt. In contrast, Niu *et al.*^[16] reported no common anastomosis of draining veins for concomitant sacral DAVF and conus medullaris AVM in a single patient successfully treated by surgery. They assumed that the formation of sacral AVF result from venous

hypertension caused by conus AVM. Recently, Guédon *et al.*^[4] reported the lumbosacral shunt developed 5 years after endovascular treatment of the conus AVF in young patient. They emphasized the anatomical and pathophysiological link that exists between conus medullaris AVF that drain caudally toward the lumbosacral area with secondary development of an acquired epidural or dural shunt. Furthermore, the focal venous hypertension and subsequent thrombosis of the main draining vein of the primary shunt after endovascular treatment can be considered responsible for local release of angiogenic factors contributing to the creation of the *de novo* fistula.

In the present study, the cauda equina AVF and sacral DAVF linked a common medullary venous channel. We speculated that primary rAVF may promote medullary venous congestion and subsequent stagnation or thrombosis inducing of a secondary sacral DAVF.

Management of the concurrent different spinal vascular malformations

The treatment of multifocal different spinal vascular malformations coexisting in the same patient remains unclear and usually difficult, especially in two lesions with distance locations. The lesion responsible for the clinical symptoms needs to be identified and treated first. However, in case of both lesions were possibly responsible for the clinical symptoms; the treatment of both lesions in the same session may be the best choice.^[6,7,16] The combination of surgical and endovascular treatments may provide a solution to treat concomitant lesions with distance locations.^[9]

Rarely, spontaneous regression of sacral DAVF occurred following embolization of conus AVF due to the thrombosis of the venous system shared by both pial and dural lesions. However, this strategy may be ineffective, and the patient may develop worsening symptoms from the other lesions left untreated.^[19]

Pitfalls of diagnostic studies

It is commonly accepted that spinal angiography is complete when one fistula has been discovered and the ASA has been visualized. However, this concept may not be sufficient in all patients. The extent and direction of the venous drainage visualized by MRI and spinal angiography should be evaluated for discrepancies.^[18]

In the present study, we found three pitfalls of diagnostic studies. First, we missed a concurrent sacral DAVF on initial spinal angiography after discovering the first fistula. Secondly, the presence of intradural hypointense vessel or flow voids in lumbar region or below the conus medullaris on T2-weighted MRI should be considered as a clue for awareness of a possible sacral DAVF. Even though we found one fistula at the level of L2, but we could not identify the venous drainage below

the conus medullaris seen on MRI. Therefore, we should keep in mind to find another possible fistula even extremely rare condition. Another last pitfall, selective catheterization of the internal iliac arteries is imperative for confirming the fistula in the sacral region supplied by the LSA. The fistula usually cannot be identified with flush aortography or selective injection of the common iliac arteries.^[3,8]

Some authors suggested that complete spinal angiography of all intersegmental arteries from the craniocervical junction to sacrum should be performed to clear of concurrent AVF.^[20] However, complete angiography needs high dose of contrast media and more radiation exposure. Contrast-enhanced spinal MRA, as shown in our case, may play an important role in the screening and detection of SDAVF before invasive angiography. MRA performed before spinal angiography and including the lumbosacral area is a valuable help to diagnose associated fistula.^[4] Therefore, spinal angiography will be focused on the level suspected the dural shunt by MRA.

After treatment one spinal vascular lesion, if the clinical status of patient fails to improve or deteriorates further with persistent flow voids seen on postoperative MR images, the possibility of reopening of the lesion or the presence of an associated malformations at different spinal level should be considered and repeat complete angiography must be performed, especially if the initial angiography was incomplete.^[2,20]

CONCLUSION

The presence of different spinal vascular malformations in a same patient is extremely rare. However, the clinician should be aware of the possibility of this condition. From our review, most authors of several studies hypothesized that venous hypertension and thrombosis due to the presence or treatment of the first spinal vascular lesion may produce a second DAVF.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Anson JA, Spetzler RF. Classification of spinal arteriovenous malformations and implications for treatment. *BNI Q* 1992;8:2-8.
2. Dam-Hieu P, Mineo JF, Bostan A, Nonent M, Besson G.

- Concurrent spinal dural and intradural arteriovenous fistulas. Case report. *J Neurosurg* 2001;95 Suppl 1:96-9.
3. Gioppo A, Faragò G, Giannitto C, Caputi L, Saladino A, Acerbi F, *et al.* Sacral dural arteriovenous fistulas: A diagnostic and therapeutic challenge-single-centre experience of 13 cases and review of the literature. *J Neurointerv Surg* 2018;10:415-21.
4. Guédon A, Condette-Auliac S, Consoli A, di Maria F, Coskun O, Rodesch G. Primary conus medullaris arteriovenous shunt and secondary lumbo-sacral epidural arteriovenous fistula: One malformation can hide another. *J Neuroradiol* 2021;48:16-20.
5. Hong T, Park JE, Ling F, terBrugge KG, Tymianski M, Zhang HQ, *et al.* Comparison of 3 different types of spinal arteriovenous shunts below the conus in clinical presentation, radiologic findings, and outcomes. *AJNR Am J Neuroradiol* 2017;38:403-9.
6. Hsu SW, Rodesch G, Luo CB, Chen YL, Alvarez H, Lasjaunias PL. Concomitant conus medullaris arteriovenous malformation and sacral dural arteriovenous fistula of the filum terminale. *Interv Neuroradiol* 2002;8:47-53.
7. Krings T, Coenen VA, Weinzierl M, Reinges MH, Mull M, Thron A, *et al.* Spinal dural arteriovenous fistula associated with a spinal perimedullary fistula: Case report. *J Neurosurg Spine* 2006;4:241-5.
8. Larsen DW, Halbach VV, Teitelbaum GP, McDougall CG, Higashida RT, Dowd CF, *et al.* Spinal dural arteriovenous fistulas supplied by branches of the internal iliac arteries. *Surg Neurol* 1995;43:35-40; discussion 40-1.
9. Li J, Li G, Bian L, Hong T, Yu J, Zhang H, *et al.* Concomitant lumbosacral perimedullary arteriovenous fistula and spinal dural arteriovenous fistula. *World Neurosurg* 2017;105:1041.e7-14.
10. Ling JC, Agid R, Nakano S, Souza MP, Reintamm G, Terbrugge KG. Metachronous multiplicity of spinal cord arteriovenous fistula and spinal dural AVF in a patient with hereditary haemorrhagic telangiectasia. *Interv Neuroradiol* 2005;11:79-82.
11. Matsumaru Y, Pongpech S, Laothamas J, Alvarez H, Rodesch G, Lasjaunias P. Multifocal and metameric spinal cord arteriovenous malformations. Review of 19 cases. *Interv Neuroradiol* 1999;5:27-34.
12. Morgalla MH, Ernemann U, Gawlowski J, Deininger M, Bitzer M, Grote EH. Recurrent spinal fistula as result of a rare combination of a perimedullary and peridural spinal fistula: Case report. *Surg Neurol* 2004;61:347-52.
13. Namba K, Niimi Y, Ishiguro T, Higaki A, Toma N, Komiyama M. Cauda equina and filum terminale arteriovenous fistulas: Anatomic and radiographic features. *AJNR Am J Neuroradiol* 2020;41:2166-70.
14. Namba K. Vascular anatomy of the cauda equina and its implication on the vascular lesions in the caudal spinal structure. *Neurol Med Chir (Tokyo)* 2016;56:310-6.
15. Nishio A, Ohata K, Takami T, Gotoh T, Tsuyuguchi N, Ichinose T, *et al.* Spinal arteriovenous malformation associated with a radicular arteriovenous fistula suggested a metameric disease. A case report. *Interv Neuroradiol* 2003;9:75-8.
16. Niu X, Ren Y, Li J. Concomitant sacral dural arteriovenous fistula and conus medullaris arteriovenous malformation with respective drainage veins: Case report and literature review.

- World Neurosurg 2020;141:299-305.
17. Ohtonari T, Ota S, Nishihara N, Suwa K, Ota T, Sekihara Y, *et al.* Arteriovenous fistula in a nerve root of the cauda equina fed by a proximal radiculo-medullary artery: A report of two cases. *Interv Neuroradiol* 2011;17:217-23.
 18. Pierot L, Vlachopoulos T, Attal N, Martin N, Bert S, Chiras J. Double spinal dural arteriovenous fistulas: Report of two cases. *AJNR Am J Neuroradiol* 1993;14:1109-12.
 19. Rosi A, Consoli A, Condette-Auliac S, Coskun O, di Maria F, Rodesch G. Concomitant conus medullaris arteriovenous shunts and sacral dural arteriovenous fistulas: Pathophysiological links related to the venous drainage of the lesions in a series of five cases. *J Neurointerv Surg* 2018;10:586-92.
 20. Sasaki O, Yajima N, Ichikawa A, Yamashita S, Nakamura K. Deterioration after surgical treatment of spinal dural arteriovenous fistula associated with spinal perimedullary fistula. *Neurol Med Chir (Tokyo)* 2012;52:516-20.
 21. Spetzler RF, Detwiler PW, Riina HA, Porter RW. Modified classification of spinal cord vascular lesions. *J Neurosurg* 2002;96 Suppl 2:145-56.
 22. Tanioka S, Toma N, Sakaida H, Umeda Y, Suzuki H. A case of arteriovenous fistula of the cauda equina fed by the proximal radicular artery: Anatomical features and treatment precautions. *Eur Spine J* 2018;27 Suppl 3:281-6.

How to cite this article: Iampreechakul P, Lertbutsayanukul P, Siriwinonmas S. Cauda equina arteriovenous fistula supplied by proximal radicular artery and concomitant sacral dural arteriovenous fistula: A case report and literature review. *Surg Neurol Int* 2021;12:405.