

Acquired Lumbosacral Spinal Dural Arteriovenous Fistula in Association with Degenerative Lumbosacral Disc Herniation and Spinal Canal Stenosis: Report of Two Cases and Review of the Literature

Abstract

The authors describe two cases harboring lumbosacral spinal dural arteriovenous fistulas (SDAVFs) manifested with nonspecific initial symptoms, leading to misdiagnosis and unnecessary procedures. A curvilinear flow void in the lumbar region and thoracic cord congestion with subtle perimedullary flow voids were detected on magnetic resonance imaging (MRI) in both patients. Contrast-enhanced magnetic resonance angiography and spinal angiography confirmed the SDAVFs in the lower lumbar and sacral region. Both fistulas were located at the same level of disc herniation and spinal canal stenosis and supplied by branches of the internal iliac arteries (i.e., iliolumbar and lateral sacral arteries) with cranial drainage from the dilated vein of the filum terminale, corresponding to a curvilinear flow void, to the perimedullary veins. The first case was successfully treated with embolization. Another case had recanalization of the fistula 4 months after endovascular treatment and was successfully treated with surgical interruption of the fistula. Our two case reports may provide additional evidence supporting an acquired etiology of SDAVFs, probably secondary to lumbosacral disc herniation and spinal canal stenosis. The authors also reviewed literature about preexistent lumbosacral SDAVFs associated with disc herniation and spinal canal stenosis. From our review, the level of SDAVFs in most patients is correlated with the level of disc herniation, spondylolisthesis, and/or spinal stenosis.

Keywords: *Acquired spinal dural arteriovenous fistula, degenerative lumbosacral spine, disc herniation, spinal canal stenosis*

Introduction

Spinal cord arteriovenous 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 (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.^[1,2]

SDAVFs are the most common type of spinal vascular malformations.^[3] These

fistulas, also known as intradural dorsal AVFs, are the arteriovenous shunts supplied by radiculomeningeal arteries draining into the radicular veins arterializing the coronal venous plexus around the spinal cord, usually under low pressure, resulting in venous congestion.^[4-6] These fistulas affect primarily the lower thoracic and upper lumbar spine and are located within the dorsal surface of the dural root sleeve in the intervertebral foramen.^[7,8] SDAVFs at the level of lower lumbar (L3–L5) or sacral region are uncommon.^[9] The patients harboring SDAVFs, typically middle-aged men, commonly manifest with progressive myelopathy induced by chronic venous hypertension.^[10] Hemorrhage from SDAVFs is usually rare and may occur as subarachnoid hemorrhage or intramedullary hemorrhage.^[11] The characteristic findings

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on T2-weighted sequences in magnetic resonance imaging (MRI) are a combination of spinal cord edema and perimedullary flow voids.^[12] SDAVFs are often initially misdiagnosed because of its insidious onset.^[13]

The pathogenesis of SDAVFs remains unclear. However, the symptoms of these fistulas usually appear in adulthood, probably considering as acquired origin.^[9] Most of SDAVFs are spontaneous. Some studies reported SDAVFs associated with trauma or previous operation.^[14-18] The authors described two cases of SDAVFs coexisting with disc herniation and spinal canal stenosis at the same level, highly suspected of a pathophysiological link. The mechanism of the formation of these fistulas in this situation was discussed.

Case Reports

Case 1

A 46-year-old male was admitted to our institute due to a history of progressive paraparesis for 2 weeks. Eight months earlier, he experienced severe left buttock pain radiating to left leg for 2 weeks. He was admitted to other private hospital and underwent discectomy L5-S1. Five months later, he suffered from right leg pain and urinary retention. He went back to the same private hospital and underwent revised surgery with spinal fusion L5-S1. Two weeks before hospitalization, he developed progressive paraparesis and constipation. He was unable to walk more than about 100 m. The patient went to another private hospital and was transferred to Prasat Neurological Institute for further investigation and proper treatment after performing MRI of the lumbosacral and thoracic spine. The neurological examination revealed the evidence of spastic paraparesis (muscle strength 3/5) and the lack of pinprick sensation below T10 level, impairment of proprioception, hyperreflexia, and presence of Babinski's sign in the lower extremities. The previous series of MRI of the lumbosacral and thoracic spine were retrospectively reviewed and revealed spinal canal stenosis and bilateral lateral recesses stenosis at L5-S1 level due to moderate diffuse annular disc bulging with moderate enlargement of facet joints and thickening of the ligamentum flavum [Figure 1]. There was an abnormal hyperintense T2 signal with

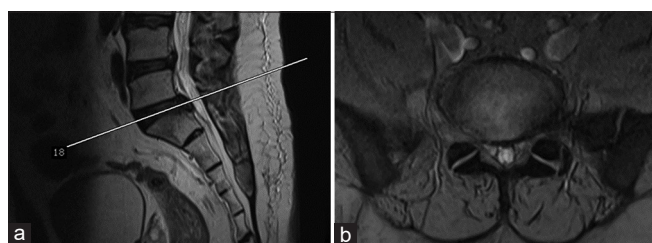


Figure 1: (a) Sagittal T2-weighted image of the lumbosacral spine and (b) axial T2-weighted image of the L5-S1 level demonstrate spinal canal stenosis and bilateral lateral recesses stenosis due to moderate diffuse annular disc bulging with moderate enlargement of facet joints and thickening of the ligamentum flavum

patchy enhancement representing spinal cord congestion extending from the conus medullaris to the level of T4 with subtle perimedullary flow voids along ventral and dorsal cord surfaces [Figures 2 and 3a]. Magnetic resonance (MR) myelography also showed “worm-like” appearance of the area from thoracic level to the conus medullaris [Figure 3b]. MR angiography (MRA) of the thoracolumbar spine demonstrated early opacification of perimedullary venous plexus along anterior and posterior surfaces of spinal cord [Figure 3c]. Furthermore, there was a curvilinear flow void, probably the dilated vein of the filum terminale, running from lower lumbar level to the conus medullaris [Figure 2]. Spinal angiography demonstrated SDAVF at the level of L5-S1, probably located on the left S1 nerve root sleeve, which is supplied by the left lateral sacral artery (LSA) originating from the internal iliac artery with venous drainage through the dilated radicular vein into the dilated vein of the filum terminale [Figure 4a and b]. The artery of Adamkiewicz arose from the left L2 lumbar artery and left T9 intercostal artery. Transarterial embolization with N-butyl-cyanoacrylate (NBCA) through the left LSA was successfully performed with NBCA filling across the fistula into the proximal draining vein [Figure 4c]. The patient had gradually improved until being the ability to walk independently 3 months later. His bowel and bladder controls had completely regained. Six months after the embolization, follow-up MRI of the thoracolumbar spine also confirmed completely regression of spinal cord congestion [Figure 5].

Case 2

A 51-year-old male, with a medical history of hypertension and diabetes mellitus, was admitted to the local hospital due to intermittent acute exacerbation of myelopathy for 3 months. One year earlier, he had developed gait

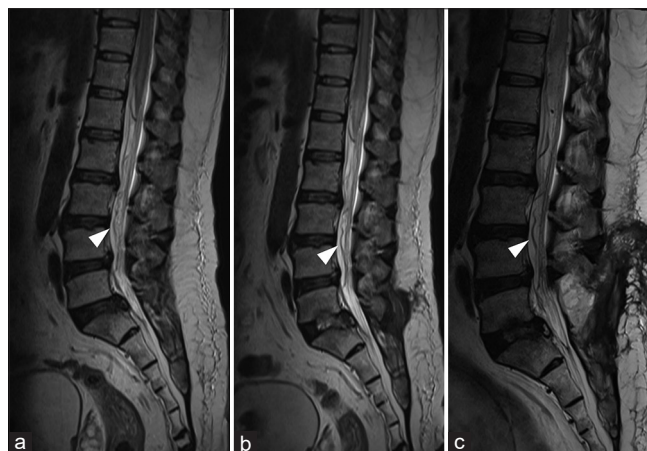


Figure 2: The series of sagittal T2-weighted magnetic resonance images of the lumbosacral spine, obtained (a) before surgery, (b) after discectomy, and (c) after spinal fusion, reveal abnormal hyperintense T2 signal representing spinal cord congestion extending from the conus medullaris to the lower thoracic. There was a curvilinear intradural flow void (arrowheads), probably the dilated vein of the filum terminale, running from the lower lumbar area to the conus medullaris

disturbance and difficulty in balancing. There was no history of trauma. Nine months later, the patient had acute onset of paraparesis after waking up in the morning. He collapsed on the ground and could not stand up for a while. After 30 min, he could raise his legs and walk as baseline. He complained of minimal back pain and denied numbness. Two months before hospitalization, he could not stand up after having dinner in the evening. After 30 min later, his symptoms improved. He had occasionally numbness on his left leg. After this event, he had a limitation on ambulation and was unable to walk more than about 50–100 m without stopping to rest. No bowel or bladder involvement was complained. Three weeks before admission, the patient had the same symptoms and rest for about 30 min then improved. MRI of whole spine was performed and was interpreted of a long segment T2 hyperintense cord lesion extending from the level of T4 to the conus medullaris. Lumbar puncture was performed due to the putative diagnosis of transverse myelitis. Cerebrospinal fluid pressure, cell count, and chemistries were within normal limits. The patient was transferred to Prasat Neurological Institute for definite diagnosis and proper management. The physical examination revealed bilateral lower extremity weakness (muscle strength 4/5) without paresthesia. Hyperreflexia was noted in the lower extremities. Babinski's sign was also positive. By counting vertebrae from whole spine screening, there was a degenerative bulging disc at the level of L4–L5 with spinal canal stenosis and compression of bilateral L5 traversing roots [Figure 6].



Figure 3: (a) Sagittal T2-weighted image of the thoracic spine shows spinal cord congestion extending from the conus medullaris to the level of T4 with subtle perimedullary flow voids along ventral and dorsal cord surfaces. (b) Coronal magnetic resonance myelography of the lumbosacral spine demonstrates "worm-like" appearance, representing a contiguous dilated and tortuous medullary vein from the conus medullaris to thoracic level. There is a dilated intradural vessel (black arrowheads) located in rather midline, probably the vein of the filum terminale. (c) Sagittal source image of contrast-enhanced spinal magnetic resonance angiography of the thoracolumbar spine reveals early opacification of perimedullary venous plexus (white arrowheads) along anterior and posterior surfaces of spinal cord

MRI of the thoracolumbar spine showed the spinal cord congestion at the level of T4 to the conus medullaris with subtle perimedullary flow voids along ventral and dorsal cord surfaces [Figure 7a and b]. Furthermore, there was a curvilinear intradural flow void on T2-weighted image, identified below the conus medullaris. Contrast-enhanced MRA of the thoracolumbar spine demonstrated an enlarged intradural vessel in the midline location extending from the lower lumbar level to the conus medullaris, probably the vein of the filum terminale [Figure 7c], corresponding with the curvilinear flow void on MRI. Spinal angiography demonstrated SDAVF at the level of L4–L5, probably located on the right L5 nerve root sleeve, which is supplied by the right iliolumbar artery originating from the internal iliac artery with cranial drainage into the dilated vein of the filum terminale. The artery of Adamkiewicz arose from the right T8 intercostal artery. Endovascular treatment with NBCA through the right iliolumbar artery was successfully performed to fill in the arterial feeder and fistulous site but fail to reach the proximal vein [Figure 8c]. Postembolization angiography of the internal iliac and iliolumbar arteries revealed complete obliteration of the fistula. The patient had gradually improved and was sent back to the local hospital for further physiotherapy. Four months later, he returned and was readmitted to our institution with gait disturbance with the deterioration of clinical symptoms. Follow-up MRI and MRA of the lumbosacral spine revealed unchanged long segment of spinal cord congestion with the presence of SDAVF at the same level. One week later, the patient underwent decompressive laminectomy

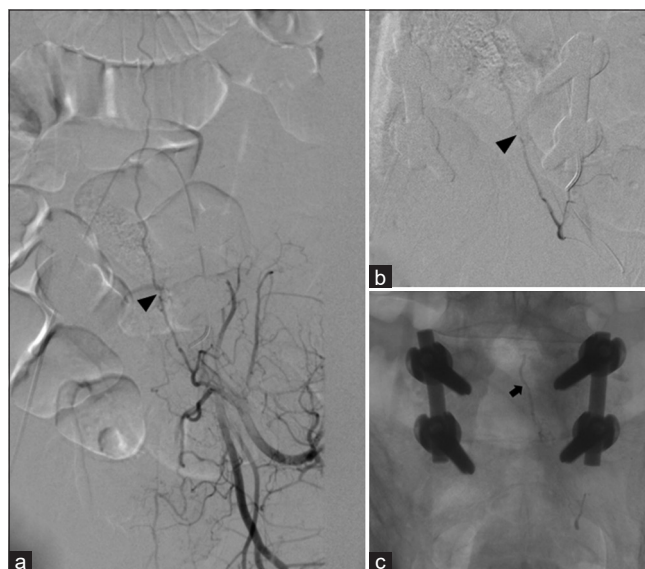


Figure 4: (a) Anteroposterior view of the left internal iliac artery angiography shows spinal dural arteriovenous fistula arrowheads, which is supplied by the left lateral sacral artery with cranial drainage into the dilated vein of the filum terminale. (b) Anteroposterior view of the left lateral sacral artery clearly demonstrates the fistula (arrowhead) at the level of L5-S1, probably located on the left S1 nerve root sleeve. (c) Anteroposterior view of nonsubtracted angiographic image reveals the glue cast (arrow) filling into the proximal draining vein



Figure 5: Sagittal T2-weighted image of the (a) lumbosacral and (b) thoracic spine, obtained six months after endovascular treatment, confirm the complete resolution of spinal cord congestion



Figure 6: (a) Sagittal T2-weighted image of the lumbosacral spine and (b) axial T2-weighted image of the L4-5 level (counting vertebral bodies from unshown whole spine screening) demonstrate spinal canal stenosis due to moderate broad base posterior bulging disc compressing bilateral L5 traversing roots



Figure 7: Sagittal T2-weighted images of the (a) lumbosacral and (b) thoracic spine show spinal congestion extending from the conus medullaris to the level of T4 with subtle perimedullary flow voids along ventral and dorsal cord surfaces. There is a curvilinear flow void (arrowheads), identified below the conus medullaris. (c) Contrast-enhanced magnetic resonance angiography of the thoracolumbar spine demonstrates an enlarged intradural vessel (arrowheads) in the midline location extending from the lower lumbar level to the conus medullaris, probably the vein of the filum terminale, corresponding with the curvilinear flow void

with obliteration of the intradural draining vein. The patient had significantly improved until being the ability to walk independently 2 weeks later. Spinal angiography, obtained 2 weeks after surgery, confirmed complete obliteration of the fistula [Figure 9]. MRI of the thoracolumbar spine, obtained 4 months after surgery, confirmed the resolution of spinal cord congestion [Figure 10].

Discussion

Delayed or missed diagnosis of SDAVFs frequently leads to unnecessary pharmacologic and/or surgical treatments and results in high rates of additional morbidity, often being irreversible despite successful obliteration of the fistula.^[19,20] A long period of venous hypertension can result in irreversible injury to spinal neurons.^[21] Based on the analysis of the patients harboring SDAVFs by Qi *et al.*,^[13] 13 patients (25%) of all the 52 patients were mistakenly diagnosed as disc herniation or lumbar spinal stenosis.

Initial symptoms of SDAVFs are often nonspecific symptoms including gait difficulties, sensory disturbances, and/or radicular pain.^[3] Patient with SDAVF associated a herniated nucleus pulposus, and spinal canal stenosis may initially present with back pain and/or radiculopathy and was initially misdiagnosed of SDAVF.^[22,23] Similarly, our first case had initial symptoms that mimic discogenic radiculopathy correlating to disc herniation and spinal canal stenosis on MRI, leading to discectomy and spinal fusion with subsequent worsening symptoms. According to a large series of 326 patients with SDAVFs by Donghai *et al.*,^[9] degenerative disc disease and myelitis were the most common misdiagnoses, and the patients were often treated incorrectly. Our second case was also misdiagnosed of myelitis due to nonspecific clinical features of intermittent acute episodes of weakness. Acute worsening of symptoms is uncommon and may occur after exercise or relate to changes in posture, probably due to an increase in venous pressure resulting from an increase in arterial pressure during physical activity.^[3]

MRI findings, including spinal cord edema or perimedullary flow voids, are important in the confirmation of the diagnosis of SDAVFs.^[12] Therefore, careful screening of MRI for conus medullaris signal or vascular flow voids is needed to avoid misdiagnosis. On myelography or MR myelography, severe spinal stenosis may display engorged and tortuous flow voids, which are possible redundant nerves and vessels, just proximal to the level of stenosis. Misinterpretation of these findings may lead to underdiagnose SDAVF, resulting in deleterious consequence. In our first case, MR myelography could identify contiguous dilated and tortuous medullary veins. Contrast-enhanced MRA may be helpful in identifying serpentine perimedullary structures and the possible arterial feeders of SDAVFs. However, spinal angiography remains the gold standard for the diagnosis.^[3]

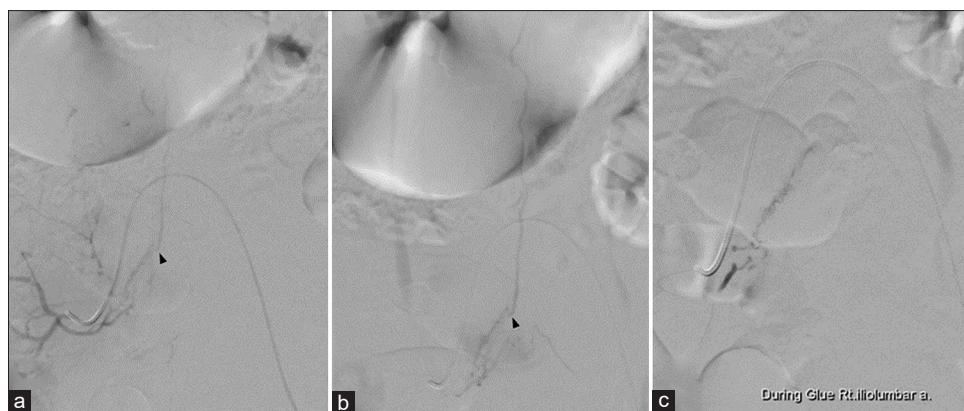


Figure 8: Anteroposterior view of the right iliolumbar artery (a) before and (b) after superselective angiography show spinal dural arteriovenous fistula (arrowheads), probably located along the right L5 nerve root sleeve, with cranial drainage into the dilated vein of the filum terminale. (c) During endovascular treatment, the glue cast fills in the distal feeder and fistulous site

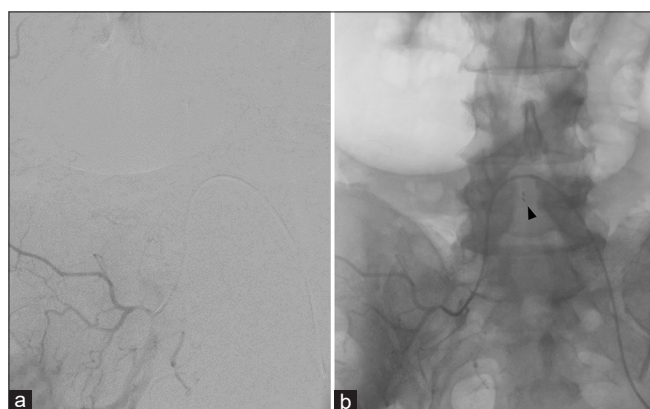


Figure 9: (a) Anteroposterior view of the right iliolumbar artery angiography, obtained 2 weeks after surgery, confirms complete obliteration of the fistula. (b) Nonsubtracted angiographic image shows small silver vessel clips (arrowhead) using during the operation

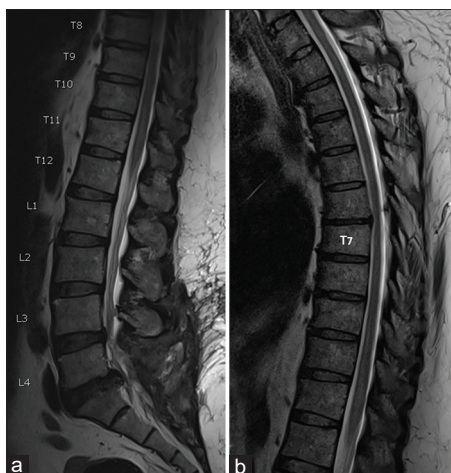


Figure 10: Sagittal T2-weighted images of the (a) lumbosacral and (b) thoracic spine, obtained 4 months after surgery, confirm the resolution of spinal cord congestion

Based on the vascular anatomy of the cauda equina reviewed by Namba,^[24] the iliolumbar artery is usually the first branch of the internal iliac artery and gives rise to a lumbar branch that travels to the intervertebral foramen

between the L5 and S1 vertebrae. Our second case, the fistula was located at the right L5 nerve root sleeve and supplied by the right iliolumbar artery. LSA, also the branch of the internal iliac artery, lies on the anterior surface of the sacral, medial to the sacral foramen. This artery gives rise to five pairs of the medial and lateral branches. The medial branches form a network of vessels and anastomose with the middle sacral artery. The lateral branches reach the anterior sacral foramen and supply the structures within the sacral canal. Our first case, the sacral DAVFs located at the left S1 root sleeve and supplied by the left LSA.

Brinjikji *et al.*^[25] studied and reviewed spinal dural and epidural AVFs and found that dural and epidural AVFs with intradural venous drainage at lower lumbar and sacral level usually drain through the vein of the filum terminale, the only intradural venous structure below the L2 vertebral body. The presence of a dilated vein of the filum terminale, identified as a curvilinear flow void on T2-weighted images, could be an imaging marker for lower lumbar and sacral fistulas. This finding can be used to facilitate to reduce radiation exposure and contrast dose. Fortunately, MRI of the lumbosacral spine in both our cases could be detected curvilinear flow voids even through perimedullary flow voids could barely be seen.

We reviewed the published case reports and series which have enough clinical description and clearly demonstrated figures of SDAVFs associated with lumbosacral disc herniation and/or spinal canal stenosis. The collected data in this review include demographic data (i.e., gender and age of patient), symptoms and signs, level of disc herniation and/or spinal canal stenosis, previous history of spinal surgery, location and artery supply of the fistulas, treatment, and neurological outcome of the patients [Table 1].^[8,16-20,22,23,26] From the literature review, there were 11 cases including our two cases. All patients were male with a median age of 54 years, range 27–79 years. The symptoms and signs of the fistulas included chronic low back pain, sciatica pain, claudication, paresthesia, gait disturbance, progressive paraparesis, and/

Table 1: Literature review of spinal dural arteriovenous fistulas associated with lumbosacral disc herniation and/or spinal canal stenosis

Authors	Sex/age	Symptoms and signs	Level of HNP/ spinal stenosis	Previous surgery	Fistula location/ side	Arterial feeders	Treatment	Outcome
Burguet <i>et al.</i> , 1985 ^[16]	Male/33	Progressive weakness and numbness of both legs, urgency of micturition	Spinal stenosis L5-S1 (probably postoperative arachnoiditis)	Yes/ discectomy	S1/R	Iliolumbar/ right, LSA/ right, left	Combined approach	GR
Yoshino <i>et al.</i> , 1998 ^[17]	Male/27	Increasing pain and numbness in the left buttock to the left lower limb	HNP L4-5, L5-S1	Yes/ discectomy, spinal fusion	L5 root/ left	Iliolumbar/ left	Combined approach	GR
Asakuno <i>et al.</i> , 2002 ^[18]	Male/60	Left-sided sciatica, progressive gait and urinary disturbances	HNP L5-S1	Yes/ discectomy	S1 root/ left	LSA/left	Surgery	GR
Stevens <i>et al.</i> , 2009 ^[22]	Male/53	Left-sided radicular symptoms, progressive loss of function in bilateral lower extremities, BBD	HNP L3-4, L4-5, L5-S1	Yes/ discectomy	L5 root/ left	L5/left, L4/right, L4/left	Embolization	PR
Nishimura <i>et al.</i> , 2014 ^[8]	Male/79	Chronic LBP with left leg numbness, progressive paraparesis, bladder dysfunction	IS L4-5, severe canal stenosis	No	L4 root/ left	L4/left	Surgery	GR (walking with cane)
Verma <i>et al.</i> , 2015 ^[23]	Male/54	Chronic LBP, new-onset left radiculopathy	HNP L1-2, L2-3, L 4-5 Spinal stenosis L1-5	Yes/ discectomy	L1-L2/ left	L1/left, L2/left	Surgery	GR
Iovtchev <i>et al.</i> , 2015 ^[20]	Male/76	Chronic LBP, difficulties in walking, spinal claudication for several months	Severe HNP L2-3, spinal stenosis L1-3	Yes/ discectomy	L1	N/A	Combined approach	PR
Brinjikji <i>et al.</i> , 2016 ^[19]	Male/68	Saddle anesthesia, numbness in the lower extremities, BBD	HNP and spinal stenosis L1-5	Yes/spinal cord biopsy	L2 root/R	L2/right	N/A	N/A
Alvarado <i>et al.</i> , 2017 ^[26]	Male/69	Long-standing LBP, progressive paraparesis, BBD	HNP and spinal stenosis L1-5	No	S1 root/R	LSA/right	Embolization	GR
Present study, 2020	Male/46	Left-sided sciatica, progressive myelopathy, BBD	HNP and spinal stenosis L5-S1	Yes	S1 root/ left	LSA/left	Embolization	GR
-	Male/51	Gait disturbance, intermittent acute episodes of weakness	HNP and spinal stenosis L4-5	No	L5 root/R	Iliolumbar/ right	Combined approach	GR

BBD – Bowel/bladder dysfunction; GR – Good recovery; HNP – Herniated nucleus pulposus; IS – Isthmic spondylolisthesis; LBP – Low back pain; LSA – Lateral sacral: Artery; N/A – Not applicable; NBCA – N-butyl cyanoacrylate; PR – Poor result

or bowel and bladder dysfunction. Eight (72.7%) patients underwent previous spine surgery. Most SDAVFs were located around or at the level of disc herniation, spinal stenosis, and/or spondylolisthesis, range L1-S1. All fistulas were supplied by lumbar artery, iliolumbar artery, and/or LSA. Surgical treatment was performed in three patients, endovascular treatment in three, and combination of surgical and endovascular treatments in four. Most patients resulted in good neurological outcome. Five of 11 patients, including our first case, had delayed or missed diagnosis of SDAVFs. ^[19,20,22,23]

The exact mechanism of the development of SDAVFs remains unclear. Acquired SDAVFs have been reported to develop secondarily to trauma. Kang *et al.*^[14] reported a 60-year-old female with the SDAVF located at the same level of a fracture L1 vertebral body. They found that all the feeding arteries, including right L1 radicular and T12

intercostal arteries, linked to the affected area and speculated that the SDAVF may result from the damage of the spinal radicular artery by fracture. Vankan *et al.*^[15] also reported the possibility of a SDAVF at the level of C1 occurring following an upper cervical spine fracture in a young male. It is likely that this traumatic fracture caused microtears of the arterial wall of the affected radiculomeningeal artery and produced an AVF. In addition, they speculated that establishing an abnormal shunt may cause by thrombosis of the perimedullary veins. Recently, Kanematsu *et al.*^[27] described a middle-aged male with an acquired cervical DAVF as a late complication after cervical anterior fusion. The radiologic findings, obtained 2 years before the initial operation, clearly showed no evidence of SDAVF. They also reviewed all previously published cases of secondary SDAVF following spinal surgery and found that some spinal epidural AVFs with or without intradural venous drainage have been recognized after lumbar surgeries.

Some reports have described a SDAVF secondary to some trauma from previous surgery. Since 1985, Burguet *et al.*^[16] reported a young male with SDAVF at the right S1 nerve root, occurred 2 years after discectomy L5-S1. The strongly increased perimedullary venous pressure in the absence of any normal epidural venous drainage is considered the main pathogenesis of clinical symptoms. They speculated that the possibility of an acquired origin of this fistula may relate to the previous discectomy, damaging epidural veins, resulting in inefficient venous drainage. Several years later, Yoshino *et al.*^[17] presented the radiographic evidence of formation of the SDAVF occurring at the same level of previous lumbar discectomy combined with spinal fusion. The investigation, obtained 7 years prior lumbar surgery, clearly showed no abnormal intradural vascular anomaly. They speculated that these operations might have induced thrombosis in the radicular vein or impairment of venous return. Asakuno *et al.*^[18] also reported a 60-year-old male with a SDAVF, developed at the site S1 nerve root sleeve damage as a result of lumbar disc extrusion. They demonstrated that MRI obtained before the discectomy showed no abnormalities. However, he had started to experience dysesthesia in both legs and progressive gait and urinary disturbances 1 year after the discectomy. MRI, obtained 2.5 years following the discectomy, revealed venous congestion extending from the conus medullaris to the level of T6. This SDAVF was supplied by the LSA. During surgical treatment for obliteration of the SDAVF, intraoperative findings demonstrated a thick fibrous scar tissue covering around an area of the fistula at the site of the previous discectomy. They speculated that an injury to the nerve root sleeve may result in the formation of this fistula.

In our literature review, seven patients, including our two cases, had preexistent fistulas coexisting with degenerative lumbosacral spondylosis.^[8,19,22,23,26] Stevens *et al.*^[22] reported a middle-aged male with a preexistent, asymptomatic SDAVF that became acutely symptomatic after lumbar discectomy. MRI of the lumbosacral spine obtained before the surgery clearly demonstrated increased signal intensity within the conus medullaris and perimedullary flow voids. Nishimura *et al.*^[8] described an elderly male with isthmic spondylolisthesis with severe canal stenosis that may give rise to the secondary SDAVF at the same level. They speculated that a SDAVF formation may develop as a consequence of any sort of trauma or inflammation associated with segmental instability inducing direct nerve root injury or impairment of venous return. Furthermore, they suggested that both the spinal dural AVF and the underlying pathology should be considered the concomitant treatment. Verma *et al.*^[23] reported a middle-aged male presented with chronic low back pain and new-onset left radiculopathy of L2. The patient underwent a left-sided L1-L2 neuroforaminal decompression and discectomy owing to a large disc extrusion causing severe compression

of the left L2 nerve root. Ten weeks after the operation, he suffered from recurrent symptoms and obtained follow-up MRI of the lumbar spine, suggesting a new disc extrusion. Due to previous extensive bleeding during previous surgery, there was concern for a vascular etiology. Therefore, spinal angiography was performed and demonstrated SDVAF at the level of L1-2, successfully treated by microsurgical resection with good results. Brinjikji *et al.*^[19] clearly demonstrated SDAVF, supplied by L2 radiculomeningeal artery, associated with severe spinal stenosis L1-5 in a 68-year-old male. Alvarado *et al.*^[26] reported sacral DAVF in a 69-year-old male presenting with long-standing low back pain and acute exacerbation of myelopathy 2 years before presentation. MRI of the lumbosacral spine clearly demonstrated severe spinal canal stenosis L3-5.

Previously, we published and reviewed the studies about acquired FTAVFs in association with degenerative lumbosacral spinal canal stenosis.^[28,29] We found that the FTAVFs were located around or at the level of spinal canal stenosis and speculated that severe spinal canal stenosis may induce local injury or inflammation to the filum terminale and produce fistulous formation. In addition, the formation of the fistula may be induced from blocked or impaired venous drainage owing to severe spinal canal stenosis. Similarly, it seems plausible that disc herniation and spinal stenosis, occurring at the same level of the fistula, are environmental factors leading to local injury to affected nerve root sleeve and/or impairment of venous outflow, resulting in the formation of SDAVF in the present study.

SDAVFs can be treated by surgery, endovascular treatment, or combined approach depends on institutions preference.^[4,5,13,18,26] The goal of treatment of SDAVF is to obliterate the fistula, including the proximal portion of the draining vein, without excision of the dilated venous plexus.^[6] Endovascular treatment has a chance of recanalization or recurrence of the fistula more than surgery.^[10,13,30] Despite more invasive technique, surgical interruption of the intradural proximal draining vein is a promising method for the treatment of the SDAVFS without recurrence.^[10] A favorable result could be achieved by surgical treatment before endovascular treatment, especially in patients with poor functional status or micturition dysregulation at diagnosis.^[21]

The potential for functional outcome was poor despite successful obliteration of the fistula and prolonged rehabilitation treatment in late diagnosis of SDAVF. Therefore, early diagnosis of SDAVF and promptly intervention treatment may result in better prognosis.^[20] SDAVFs associated with lumbosacral disc herniation and/or spinal canal stenosis should be suspected when the patients develop progressive myelopathy and/or bowel/bladder dysfunction. MRI scans should be inspected carefully for the evidence of the fistula, including increased

T2 hyperintensity in spinal cord from the conus medullaris extending up to thoracic level, abnormal curvilinear intradural flow void, and/or perimedullary flow voids. Spinal MRA and/or angiography may be required to confirm this possibility.

Conclusion

SDAVFs are widely accepted to be an acquired in origin due to symptomatic onset in adulthood. The exact etiology of SDAVFs remains unclear. SDAVFs at the level of lower lumbar or sacral region are rare. It has been reported that some trauma from the previous surgery resulted in the development of these diseases. Our two case reports may provide additional evidence supporting an acquired etiology of SDAVFs, probably secondary to lumbosacral disc herniation and spinal canal stenosis. From our review, the level of SDAVFs in most patients is correlated with the level of disc herniation, spondylolisthesis, and/or spinal canal stenosis. It seems plausible that disc herniation and spinal stenosis are environmental factors leading to local injury to affected nerve root sleeve and/or impairment of venous outflow, resulting in the formation of SDAVF. It should be aware of SDAVF when the patients have disc herniation and/or spinal stenosis.

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 understood that name 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.

References

1. Anson JA, Spetzler RF: Classification of spinal arteriovenous malformations and implications for treatment. *BNI Quarterly* 1992;8:2-8.
2. Barrow DL, Colohan AR, Dawson R. Intradural perimedullary arteriovenous fistulas (type IV spinal cord arteriovenous malformations). *J Neurosurg* 1994;81:221-9.
3. Jellema K, Tijssen CC, van Gijn J. Spinal dural arteriovenous fistulas: A congestive myelopathy that initially mimics a peripheral nerve disorder. *Brain* 2006;129:3150-64.
4. Spetzler RF, Detwiler PW, Riina HA, Porter RW. Modified classification of spinal cord vascular lesions. *J Neurosurg* 2002;96:145-56.
5. Krings T, Thron AK, Geibprasert S, Agid R, Hans FJ, Lasjaunias PL, *et al.* Endovascular management of spinal vascular malformations. *Neurosurg Rev* 2010;33:1-9.
6. Anson JA, Spetzler RF. Interventional neuroradiology for spinal pathology. *Clin Neurosurg* 1992;39:388-417.
7. Rosenblum B, Oldfield EH, Doppman JL, Di Chiro G. Spinal arteriovenous malformations: A comparison of dural arteriovenous fistulas and intradural AVM's in 81 patients. *J Neurosurg* 1987;67:795-802.
8. Nishimura Y, Natsume A, Ginsberg HJ. Spinal dural arteriovenous fistula associated with L-4 isthmic spondylolisthesis. *J Neurosurg Spine* 2014;20:670-4.
9. Donghai W, Ning Y, Peng Z, Shuo X, Xueen L, Peng Z, *et al.* The diagnosis of spinal dural arteriovenous fistulas. *Spine (Phila Pa 1976)* 2013;38:E546-53.
10. Kiyosue H, Matsumaru Y, Niimi Y, Takai K, Ishiguro T, Hiramatsu M, *et al.* Spinal AV Shunts Study Group. Angiographic and clinical characteristics of thoracolumbar spinal epidural and dural arteriovenous fistulas. *Stroke* 2017;48:3215-22.
11. Iampreechakul P, Lertbutsayanukul P, Siriwiwommas S, Jittapiromsak P, Tantivatana J, Niruthisard S. Persistent central neuropathic pain caused by intramedullary hemorrhage from spinal dural arteriovenous fistula: A case report and literature review. *Austin J Anesthesia Analgesia* 2019;7:1076.
12. Jeng Y, Chen DY, Hsu HL, Huang YL, Chen CJ, Tseng YC. Spinal dural arteriovenous fistula: Imaging features and its mimics. *Korean J Radiol* 2015;16:1119-31.
13. Qi X, Lv L, Han K, Xu Z, Mei Q, Chen H, *et al.* Analysis of the embolization spinal dural arteriovenous fistula and surgical treatments on 52 cases of the patients. *Int J Clin Exp Med* 2014;7:3062-71.
14. Kang JW, Koo JH, Kim DK, Joo YJ, Kim TH, Song SH. Myelopathy caused by spinal dural arterio-venous fistula after first lumbar vertebral body fracture - a case report -. *Ann Rehabil Med* 2011;35:729-32.
15. Vankan Y, Demaerel P, Heye S, Van Calenbergh F, van Loon J, Maleux G, *et al.* Dural arteriovenous fistula as a late complication of upper cervical spine fracture. Case report. *J Neurosurg* 2004;100:382-4.
16. Burguet JL, Dietemann JL, Wackenheim A, Kehr P, Buchheit F. Sacral meningeal arteriovenous fistula fed by branches of the hypogastric arteries and drained through medullary veins. *Neuroradiology* 1985;27:232-7.
17. Yoshino O, Matsui H, Hirano N, Tsuji H. Acquired dural arteriovenous malformations of the lumbar spine: Case report. *Neurosurgery* 1998;42:1387-9.
18. Asakuno K, Kim P, Kawamoto T, Ogino M. Dural arteriovenous fistula and progressive conus medullaris syndrome as complications of lumbar discectomy. Case report. *J Neurosurg* 2002;97:375-9.
19. Brinjikji W, Nasr DM, Morris JM, Rabinstein AA, Lanzino G. Clinical Outcomes of Patients with Delayed Diagnosis of Spinal Dural Arteriovenous Fistulas. *AJNR Am J Neuroradiol* 2016;37:380-6.
20. Iovtchev I, Hiller N, Ofra Y, Schwartz I, Cohen J, Rubin SA, *et al.* Late diagnosis of spinal dural arteriovenous fistulas resulting in severe lower-extremity weakness: A case series. *Spine J* 2015;15:e39-44.
21. Kang MS, Kim KH, Park JY, Kuh SU, Chin DK, Jin BH, *et al.* Comparison of Endovascular Embolization and Surgery in the Treatment of Spinal Intradural Dorsal Arteriovenous Fistulas. *World Neurosurg* 2019;122:e1519-e1527.

22. Stevens EA, Powers AK, Morris PP, Wilson JA. Occult dural arteriovenous fistula causing rapidly progressive conus medullaris syndrome and paraplegia after lumbar microdiscectomy. *Spine J* 2009;9:e8-12.
23. Verma K, Fennessy J, Huang R, Jabbour P, Rihn J. Spinal dural arteriovenous fistula presenting as a recurrent nucleus pulposus herniation: A case report and literature review. *JBJS Case Connect* 2015;5:e59.
24. 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.
25. Brinjikji W, Hilditch CA, Morris JM, Dmytriw AA, Cloft H, Pereira VM, *et al.* Dilated vein of the filum terminale on MRI: A marker for deep lumbar and sacral dural and epidural arteriovenous fistulas. *AJNR Am J Neuroradiol* 2018;39:1953-6.
26. Alvarado AM, Haussen DC, Ebersole K, Nogueira RG, Abraham MG. Embolization of sacral dural arteriovenous fistulas: A case series and literature review. *Interv Neurol* 2017;6:73-81.
27. Kanematsu R, Hanakita J, Takahashi T, Tomita Y, Minami M. An acquired cervical dural arteriovenous fistula after cervical anterior fusion: Case report and literature review. *World Neurosurg* 2019;128:50-4.
28. Iampreechakul P, Tirakotai W, Lertbutsayanukul P, Khunvutthidee S, Thammachantha S, Siriwimonmas S. Spinal sparganosis coexisting with acquired arteriovenous fistula of the filum terminale. *World Neurosurg* 2020;136:341-7.
29. Iampreechakul P, Tirakotai W, Wangtanaphat K, Lertbutsayanukul P, Siriwimonmas S. Filum terminale arteriovenous fistula in association with degenerative lumbosacral spinal canal stenosis: Report of 3 cases and review of the literature. *World Neurosurg* 2020;138:231-41.
30. Adamczyk P, Amar AP, Mack WJ, Larsen DW. Recurrence of “cured” dural arteriovenous fistulas after Onyx embolization. *Neurosurg Focus* 2012;32:E12.