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Case Report

Sacral dural arteriovenous fistula of the filum terminale coexisting with partially thrombosed filum vein: A case report and literature review

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ABSTRACT

Background: Filum terminale arteriovenous fistulas (FTAVFs) are rare and usually classified as intradural ventral AVFs or Type IVa perimedullary fistulas, located on the pia surface along the course of filum terminale internum (FTI). We report an extremely rare case of sacral dural arteriovenous fistula of the FT. We also review the occurrence of FTAVFs in the sacral region.

Case Description: A 64-year-old man presented with progressive weakness of the lower extremities for 3 months and bowel/bladder dysfunction following long history of back pain radiating to both legs. Magnetic resonance imaging of the lumbosacral and thoracic spine showed spinal cord congestion, extending from the conus medullaris to the level of T3, and partial thrombosis within the abnormal tortuous and dilated flow void, running from the sacral area to conus medullaris. Further findings were compression fracture of L2 vertebra, Grade I degenerative spondylolisthesis at the level of L2-3, and L3-4, and spinal stenosis at L2-3, L3-4, and L4-5. Spinal angiography, maximum intensity projection reformatted image of angiographic computerized tomography, and three-dimensional reconstructed image clearly demonstrated dural AVF of the FT at the level of S2 supplied by bilateral lateral sacral and middle sacral arteries with cranial drainage to perimedullary vein through the enlarged vein of the filum. The patient was indirectly treated by transection of the filum terminale and the draining vein at the level of L5 rostral to the fistula.

Conclusion: Sacral DAVFs of the FT are extremely rare. In our case, the formation of fistula may cause by venous hypertension secondary to partial thrombosis within the filum vein, probably resulting from long-standing spinal canal stenosis. Sacral FTAVFs may be found on the pia surface of the terminal FTI, dural component at the area of dural sac termination, or dural extension covering the filum terminale externum.

Keywords: Fatty filum terminale, Filum terminale arteriovenous fistula, Filum terminale externum, Sacral dural arteriovenous fistula, Thrombosis of draining vein

INTRODUCTION

Filum terminale arteriovenous fistulas (FTAVFs), located below the conus medullaris along the course of the filum, are characterized by a single direct communication between the artery

of the FT, distal termination of the anterior spinal artery (ASA), and the vein of the FT without intervening nidus.[5] FTAVFs are classified as Type IVa perimedullary fistulas, usually located against the anterior aspect of the conus medullaris or the filum terminale.[19] FTAVFs are rare and account for approximately 3% of all spinal arteriovenous lesions.[22] These fistulas often manifest with symptoms of congestive myelopathy secondary to venous hypertension and usually affect middle-aged men.[24] FTAVFs are usually located on the pia surface along the course of filum terminale internum (FTI).[29,30]

FTAVFs coexisting with spinal dysraphism, including tethered spinal cord and lipoma, may be found in the sacral region.[26] Without the low-lying spinal cord and sacral lipoma, dural AVFs (DAVFs) of the FT at the sacral region are exceedingly rare.[8]

We reported an extremely rare case of sacral DAVF of the FT coexisting with partially thrombosed filum vein. In addition, we reviewed the published case reports and series which have enough clinical description and clearly demonstrated figures of FTAVFs at the sacral region without spinal dysraphism.

CASE DESCRIPTION

A 64-year-old man was admitted to our institute due to a 3-month history of progressive paraparesis. Three years earlier, he experienced low back pain radiating to both legs, predominantly affecting the right side. He also had numbness in lower extremities, affecting the left more than right side. He was treated by medicine and physiotherapy at the local hospital. One month before the hospitalization, he was unable to walk without assistant. In addition, urine retention and constipation were noticed 2 weeks before admission. His back was injured by falling from a tree at a height of 3 m 4 years ago. The neurological examination revealed the evidence of spastic paraparesis (muscle strength 2/5), the lack of pinprick sensation below T10 level, hyperreflexia, and presence of Babinski sign in the lower extremities.

Magnetic resonance imaging (MRI) of the thoracolumbar spine showed hyperintense T1 signal, hypointense T2 signal with blooming on gradient-echo (GRE) T2*-weighted image in the abnormal flow void running from sacral level to the conus medullaris, probably representing subacute thrombosis. There were abnormal hyperintense T2 signal representing spinal cord congestion extending from the conus medullaris to the level of T3 and subtle perimedullary flow voids along the posterior surface of cord. After gadolinium contrast, diffuse enhancement of lower spinal cord and cauda equina nerve roots was observed [Figure 1]. Further findings were compression fracture of L2 vertebra, Grade I degenerative retrolisthesis at the level of L2-3, and L3-4, and thickened ligamentum flavum with hypertrophic facet joints at the level of L2-3, L3-4, and L4-5, causing spinal stenosis at L2-3, L3-4, and L4-5. Contrast-enhanced magnetic resonance angiography (MRA) of the thoracolumbar spine demonstrates tortuous and enlarged intradural vessels at the midline location extending from the level of lower lumbar to thoracic level [Figure 2].

Spinal angiography demonstrated an AVF at the level of S2, which is supplied by the branches from bilateral lateral sacral arteries (LSA) with cranial drainage into the dilated vein of the FT. The middle sacral artery (MSA) anastomosing with the distal branch of left LSA was identified. The ASA arose from the left L3 segmental artery without supplying to the fistula. Maximum intensity projection (MIP) reformatted image of angiographic computerized tomography (CT) and three-dimensional reconstructed image clearly revealed the fistulous point at the level of S2. The fistula fed by two branches from the left LSA. One upper branch ran through the fourth sacral foramen. Another lower branch traveled through the sacral hiatus and then joins the straight artery along the filum terminale externum (FTE). The fistula was supplied by the right LSA through the right first sacral foramen [Figures 3 and 4].

The patient underwent surgical treatment. To avoid invasive posterior sacrectomy, we decided to obliterate the dilated intradural draining vein rostral to the fistula. Laminotomy was performed at the level of L5. After durotomy, the arachnoid membrane was found to be thickened and opaque. The arachnoid was gently separated. The roots of the cauda equina were mattered together with adhesions. The FT was adhered within the clumping of the cauda equina. Lysis of adhesions between the nerve roots was performed. After lysis of adhesions, the engorged vein run parallel with the FT was identified. Part of the dilated vein was resected along with the FT [Figure 5]. Histopathological examination disclosed dilated vessel and thickened vascular wall, surrounded by fibrofatty tissue. Attached small nerve fibers are noted. These findings were consistent with dilated vein of the FT embedded in fatty FT [Figure 6].

His postoperative course was uneventful. Follow-up spinal angiography obtained 1 week after the operation confirmed complete obliteration of an arteriovenous fistula [Figure 7]. He was discharged home 2 weeks later and was sent to the local hospital for physical rehabilitation. MRI of the thoracolumbar spine obtained 3 months after the surgery showed hyperintense T1 and T2 signal with blooming on GRE image along the flow void running from the level of L5 to L2, probably representing complete thrombosis of the vein of the FT above the clipping point. There was only minimal persistent central cord enhancement [Figure 8]. Moreover, the resolution of spinal cord congestion and disappearance of perimedullary flow voids were observed. Contrast-enhanced MRA of the thoracolumbar spine confirmed no recurrent of the fistula [Figure 9]. At the 4-month follow-up, the patient was able to walk independently without bowel/bladder

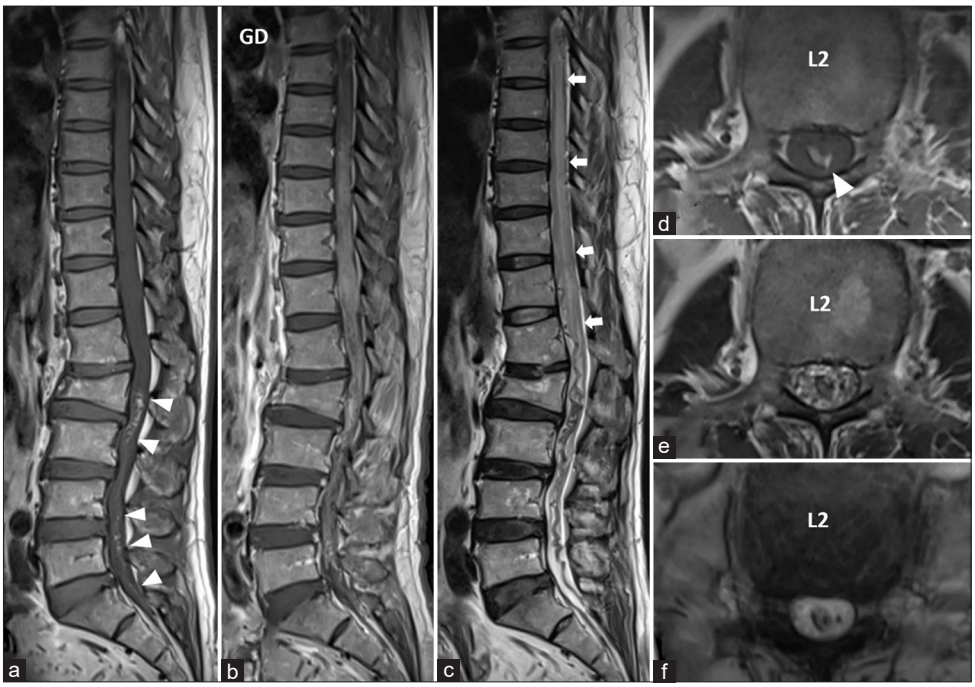

Figure 1: Preoperative magnetic resonance imaging of the thoracolumbar spine. Sagittal views of T1-weighted (a) without and (b) with gadolinium injection, (c) T2-weighted, and axial (d) T1-weighted, (e) T2-weighted, and (f) gradient-echo (GRE) T2*-weighted images at the level of L2 demonstrate hyperintense (arrowheads) T1 signal, hypointense T2 signal with blooming on GRE image in the flow void running from sacral level to the conus, probably representing subacute thrombosis. There is spinal cord congestion (arrows) extending from the conus to thoracic level. Enhancement of long segment spinal cord and cauda equina nerve roots are noted. Compression fracture of L2 vertebra is observed.

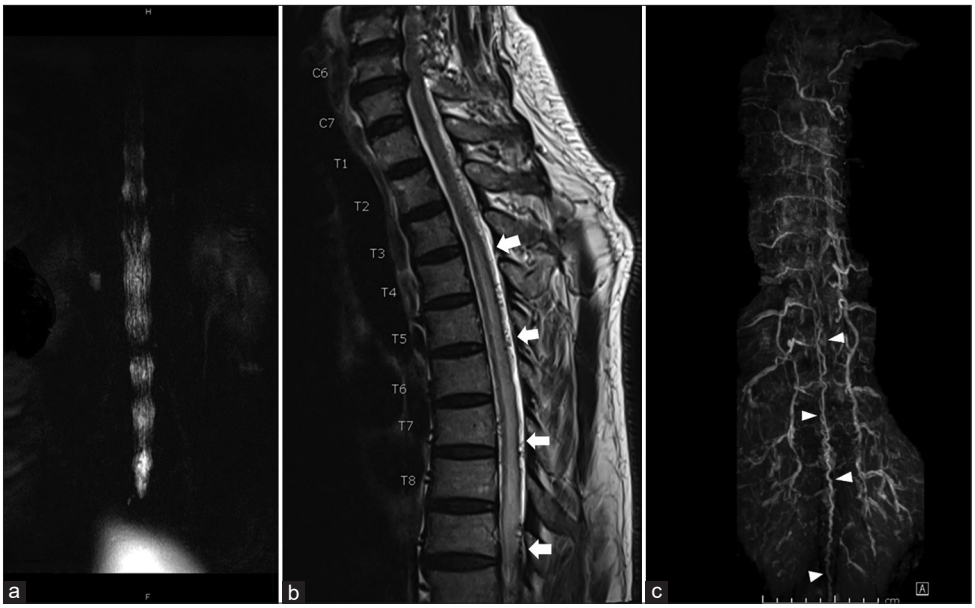

Figure 2: (a) Coronal myelographic sequence reveals spinal canal stenosis corresponding with filling defect at the level of L2–3, L3-4, and L4–5. (b) Sagittal T2-weighted image of the cervicothoracic spine shows spinal cord congestion (arrows) extending up to T3 level and subtle intradural flow voids along the posterior surface of cord. (c) 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 lower lumbar to thoracic level.

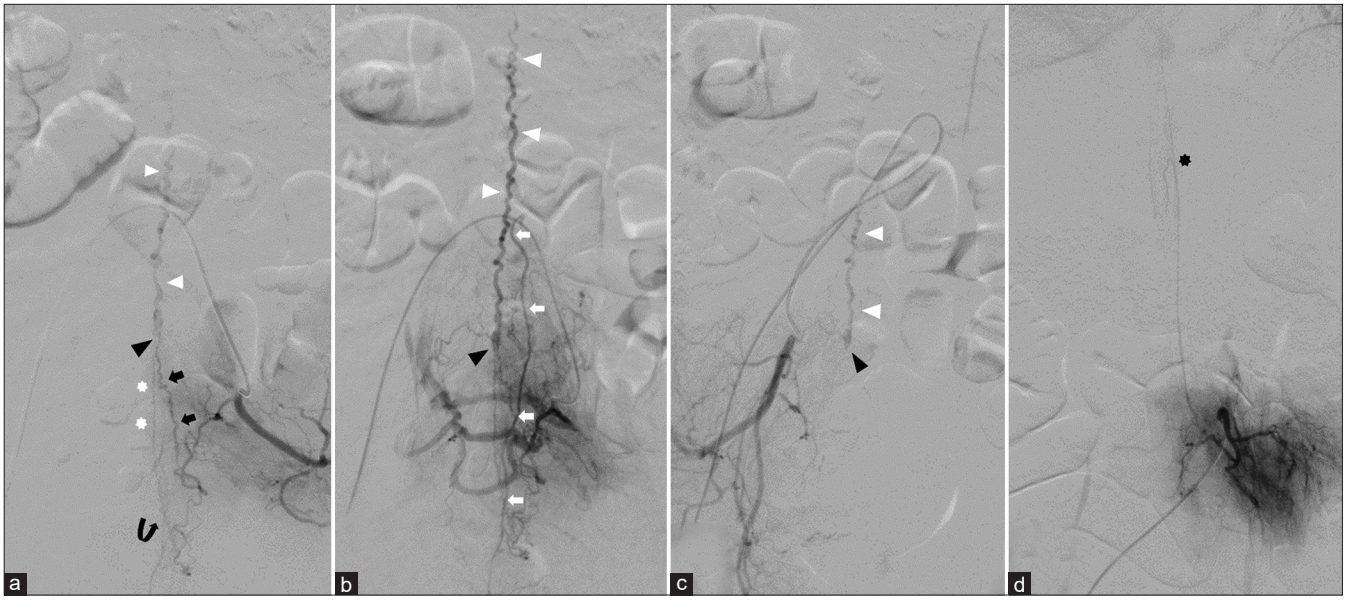

Figure 3: Preoperative spinal angiography. Arterial (a) and venous (b) phases of the left internal iliac artery injection show an arteriovenous fistula (black arrowheads) at the midline at the level of S2 supplied by the left lateral sacral artery (LSA) with cranial drainage into the dilated vein of the filum terminale (white arrowheads). There are two branches from the left LSA supplying to the fistula. One upper branch (black arrows) runs through the fourth sacral foramen. Another lower branch (curve arrow) travels through the sacral hiatus and then joins the straight artery (white asterisks) along the filum terminale externum. The middle sacral artery (white arrows) anastomosing with the distal branch of left LSA is identified. (c) The right internal iliac artery injection also reveals the same fistula (black arrowhead) with the dilated filum vein (white arrowheads) fed by the right LSA through the right first sacral foramen. (d) The left L3 segmental artery injection illustrates an anterior spinal artery with a characteristic hairpin turn (black asterisk).

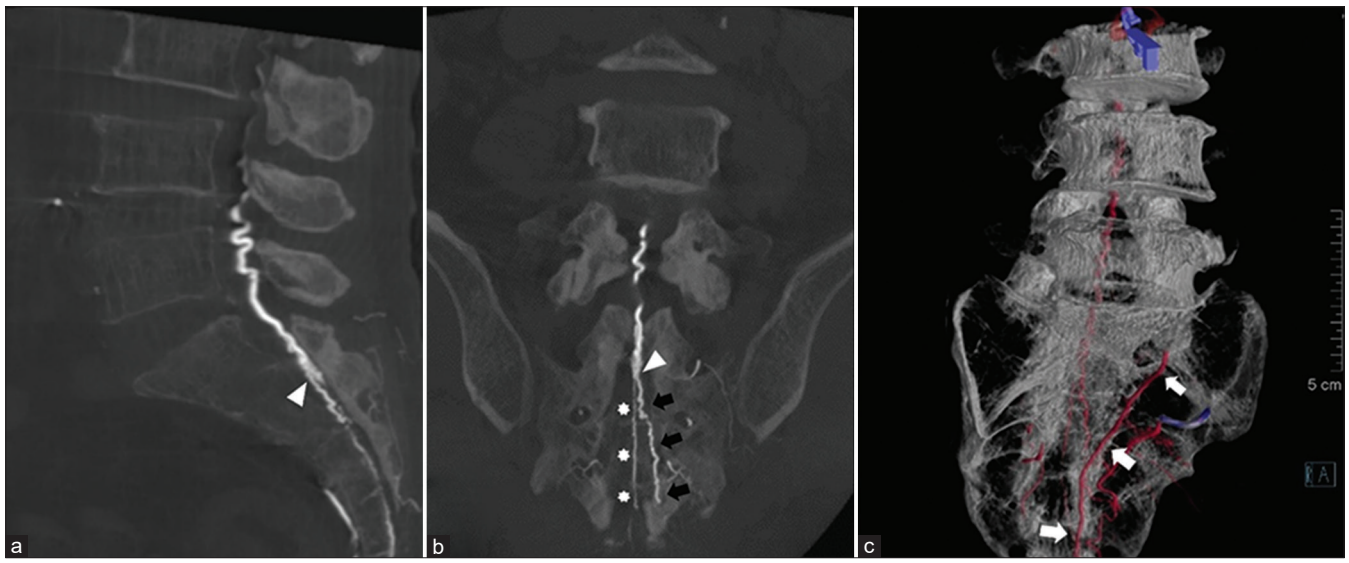

Figure 4: Sagittal (a) and coronal (b) maximum intensity projection reformatted images of angiographic computerized tomography of the left internal iliac artery clearly show the fistulous point (arrowheads) at the level of S2. The dural branch (black arrows) from the left lateral sacral artery runs through the left fourth sacral foramen and supplies to the fistula. Below the fistula, there is a straight artery (asterisks) run along the filum terminale externum at midline location. (c) Three-dimensional reconstructed image demonstrates the middle sacral artery (white arrows).

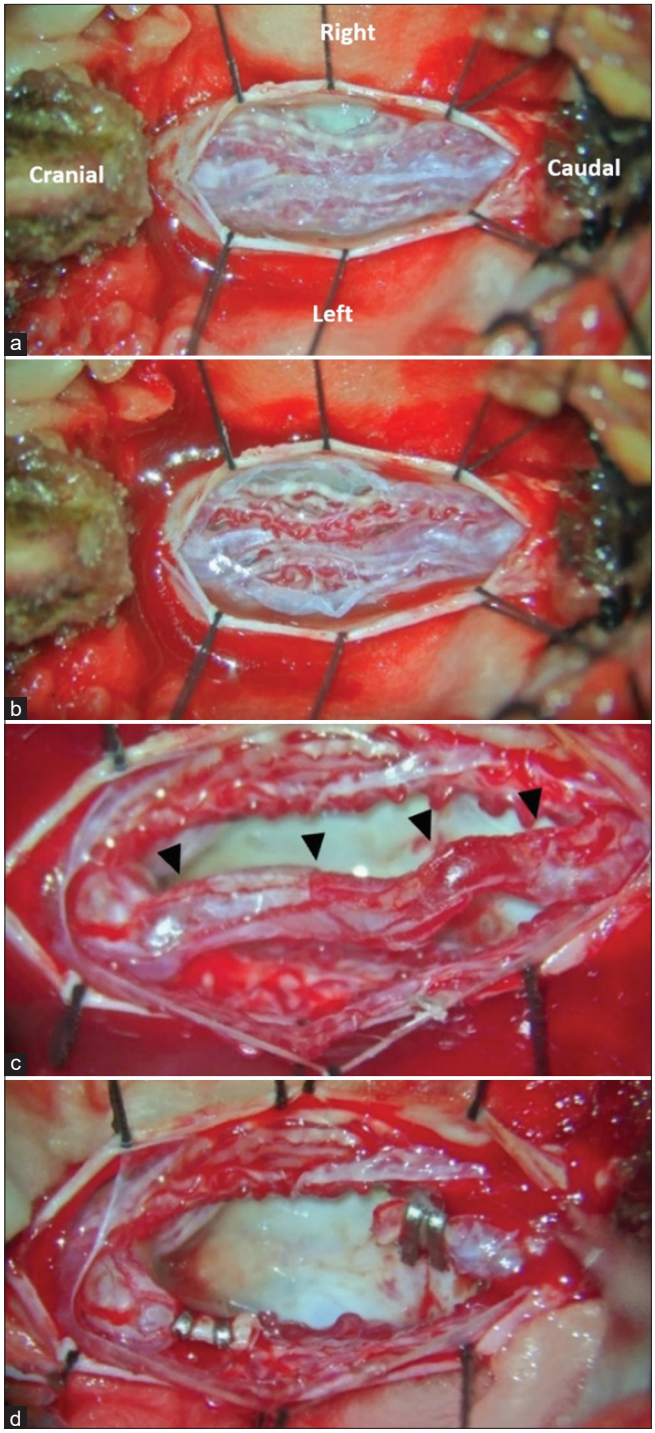

Figure 5: Intraoperative photograph during surgery on prone position after opening the dura at L5-S1 level. (a) the arachnoid membrane was found to be thickened and opaque. The roots of the cauda equina were mattered together. (b) After lysis of adhesions (c) The engorged vein run parallel with the filum terminale (arrowheads). (d) After clipping and resection of the filum terminale and dilated vein.

dysfunction, although he still experienced numbness in his left leg.

DISCUSSION

The anatomy and blood supply of the FT

The FT is a thin fibrous band that attaches the tip of the conus medullaris to the dorsal surface of the coccyx.[1] It is divided into two parts, including the FTI (intradural part), and FTE (extradural part). The FTI begins at the caudal tip of the conus medullaris extension distally and ends by fusion or piercing with the dura mater of the terminal thecal sac. The extradural part begins at the apex of the dural thecal sac and traces distally into its attachment into the dorsal surface of the coccyx.[28] According to cadaveric study of FT fusion and dural sac termination by Hansasuta *et al*.,[7] they found that the majority of the distal FTI fuses with dura mater at the level of S2 with a range from L5 to S3. In addition, the majority of dural sac termination occur at S2 with a range from S1-S3.

The artery of FT is the main feeding artery of the FTI from the tip of the conus medullaris to the area fused with the dural sac termination and can barely supply the FTE.[23] Based on the study of the vascularization of the FT by Djindjian *et al*.,[2] the artery of the FT 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 FT gives origin in arterioles supplying the coccygeal nerve roots adherent to the filum. Therefore, the vascular lesion of FT may receive additional supply from the LSA.[21]

The blood supply of the FTE remains unclear. The proximal FTE, junction with the dural sac termination, always contains remnants of meningothelial tissue.[28] Therefore, the FTE covered by dural membrane could be fed by the dural or dorsal somatic branches originating from iliolumbar, middle sacral, or lateral sacral arteries.[23,29] Regarding histological analysis in 15 adult cadavers by Tubbs *et al*.,[28] they found that straight blood vessel following the course of the FTE have been identified in ten cases. However, the origin of this vessel remains unknown. In our study, we can demonstrate the straight blood supply on the FTE using MIP reformatted image of angiographic CT. We speculated that the straight artery along the FTE may be named the artery of the FTE, probably arising from the LSA or MSA.

The MSA arises from the caudal end of the abdominal aorta at the dorsal surface. The branches of the LSA form numerous anastomoses with the lateral branches of the MSA.[21] Djindjian *et al*.[3] illustrated anastomoses between the MSA and bilateral LSAs at the lower sacral region.

The vein of the FT, always larger than the artery of the FT, is located on the ventral aspect of the filum behind the artery of the FT throughout its course on the filum. The bidirectional venous drainage of this vein includes descending towards the sacral venous plexuses and ascending toward the perimedullary veins

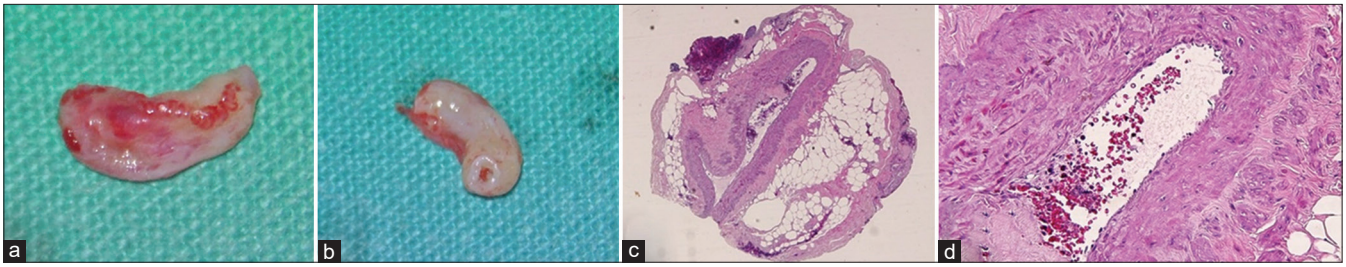

Figure 6: (a and b) Gross specimens of the resected filum terminale and dilated vein of the filum. (c and d) The histopathological study shows dilated vessel and thickened vascular wall surrounded by fibroadipose tissue. Thick vascular wall consists of inner circular and outer longitudinal layer of smooth muscles (hematoxylin and eosin ×40 and ×200).

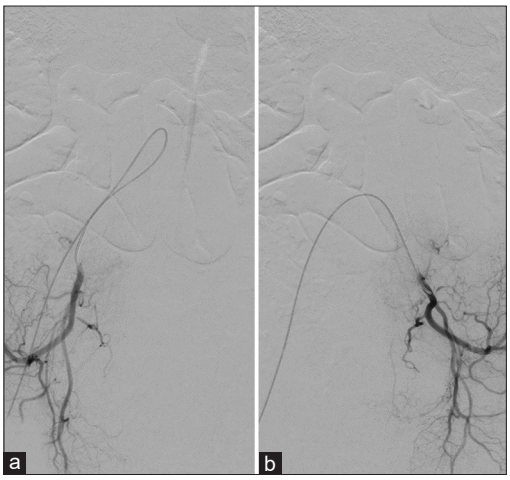

Figure 7: Spinal angiography obtained 1 week after the operation, anteroposterior views of (a) right and (b) left internal iliac arteries injections confirm complete obliteration of an arteriovenous fistula.

through anterior and posterior spinal veins or the juxtamedullary anastomoses at the level of the conus medullaris.[2]

Literature review of sacral FTAVFs without spinal dysraphism

In our literature review, the collected data in this review include demographic data (i.e., gender and age of patient), symptoms and signs, location, and artery supply of the fistulas, treatment, and neurological outcome of the patients. There were eight cases, including our one case with nine sacral FTAVFs [Table 1].[8,14,16,18,23,29,30] Seven (87.5%) men and one (12.5%) woman with median age 63 years, range 42–78 years, were included in this review. The symptoms and signs of the fistulas included back pain with or without sciatica, intermittent claudication, paresthesia, progressive myelopathy, and/or bowel/bladder dysfunction. FTAVFs were located at the distal FTI in three patients, FTE in 2, and dura mater in 4, range S1-S4. Most fistulas, including our one case, located at the FTE and dura mater were supplied mainly by the LSA and/or MSA.[8,14,16,23] Only one case of DAVF of the FT had an additional supply by the artery of the FT arising from ASA.[29] Whereas the other three fistulas located at the FTI were fed mainly by the artery of the FT.[16,18,30] Surgical treatment was performed in five patients (62.5%), endovascular treatment in 2 (25%), and combination of surgical and endovascular treatments in 1 (12.5%). Most patients resulted in good neurological outcome.

Spontaneous partial thrombosis of the vein of FT

Spontaneous thrombosis of spinal vascular malformations, including intramedullary arteriovenous malformation (AVM), perimedullary AVF, and DAVF, has been previously reported and is extremely rare. The mechanism of thrombosis remains unclear. Most thrombosed part of these lesions was the draining vein within spinal cord. No abnormal vessels on spinal angiography were identified. This condition is likely precipitate a rapid venous hypertension, leading to acute deterioration after a subacute myelopathic course, known as Foix-Alajouanine syndrome.[27] In the present study, we believed that the symptoms and image findings in our case were not consistent with Foix-Alajouanine syndrome due to progressive symptoms without acute deterioration, no thrombosis of veins within spinal cord, the presence of draining vein on angiography, and significantly improvement of the patient following surgery.

Regarding signal intensities of MRI, hyperintense changes in the flow void or draining vein on T1-weighted sequences usually represent subacute thrombosis.[6] Signal in T2-weighted image depends on the rate of residual venous flow.[4] Intraluminal thrombus is hyperintense on T2-weighted sequences, whereas flowing blood is hypointense.[31] The different MRI signal intensities of thrombus in the filum vein in our case were correlated with pre and postoperative spinal angiography. Preoperative MRI demonstrated hyperintense T1 signal and hypointense T2 signal in the flow void, representing subacute partial thrombosis in the vein of the filum with patent lumen, as shown in preoperative spinal angiography. After the surgery at 3 months, postoperative MRI revealed hyperintense T1 signal and hyperintense T2 signal, probably representing complete thrombosis of the vein of the FT from above clipping point to L2 level. Postoperative spinal angiography confirmed complete obliteration of the fistula and no flowing blood in the draining vein.

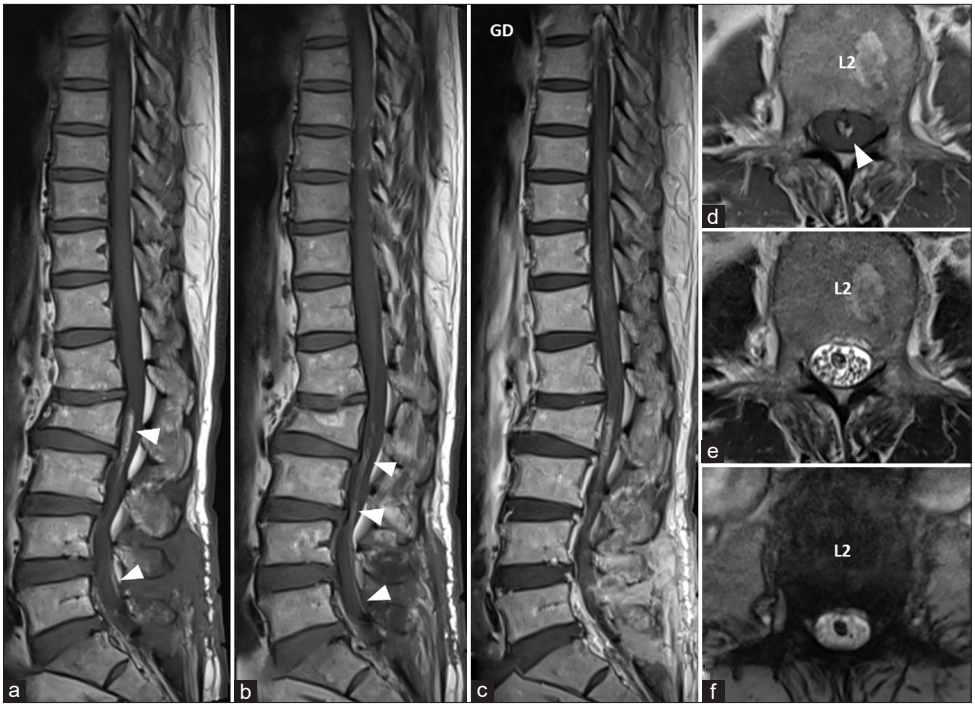

Figure 8: Magnetic resonance imaging of the thoracolumbar spine obtained 3 months after the operation. Sagittal views of T1-weighted (a and b) without and (c) with gadolinium injection and axial (d) T1-weighted, (e) T2-weighted, and (f) gradient-echo (GRE) T2*-weighted images at the level of L2 demonstrate hyperintense T1 (arrowheads) and T2 signal with blooming on GRE image along the flow void running from the level of L5 to L2, probably representing complete thrombosis of the vein of the filum terminale above the clipping point. There is only minimal persistent central cord enhancement.

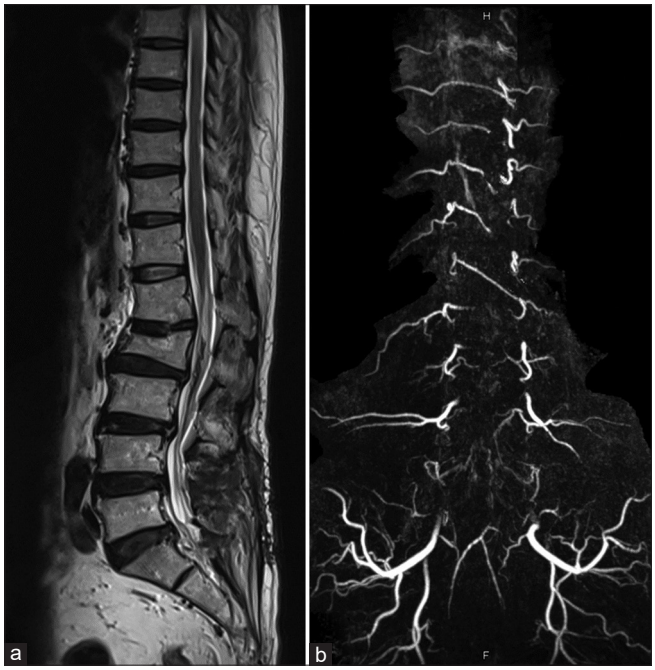

Figure 9: Sagittal T2-weighted image of the thoracolumbar spine obtained 3 months after the operation demonstrates the resolution of spinal cord congestion and disappearance of perimedullary flow voids. (b) Contrast-enhanced magnetic resonance angiography of the thoracolumbar spine confirms complete obliteration of the fistula.

Sacral DAVF of the FT

Sacral DAVF of the FT should be differentiate from cauda equina AVF, which was supplied from dural branches of the internal iliac and/or lateral sacral arteries. However, the drainage veins are radicular veins and usually follow the nerve roots, which locate more laterally and near the region of the sacral foramen.[8] The radicular vein may be misinterpreted as the filum vein. Occasionally, intraoperative findings may be needed to differentiate the radicular vein from the vein of the filum.[11]

Ryu *et al*.[23] demonstrated a case of an AVF located in the FTE at the level of S2-3. Using image fusion of MRI images with postoperative CT, they concluded that this fistula was DAVF occurring in the dural extension covering the FTE. In the present study, the dural termination occurred at the level of S2, where the location of the fistula existed. We speculated that this fistula was a dural shunt of the FT close to the dural attachment at the area of dural sac termination, which was shunted into the dilated filum vein. The dilated vein of the FT was confirmed by the pathological study. Due to the presence of the MSA during injection of the left LSA, we believed that this fistula was not only fed by the LSAs, but also supplied by the MSA. However, we did not inject the MSA for proof this expectation.

Table 1: Literature review of patients with filum terminale arteriovenous fistula at sacral region without spinal dysraphism.

Authors	Gender/Age	Symptoms and signs	Location of the fistula	Main feeder	Treatment	Neurological outcome
Hsu *et al.* (2002)[8]	M/44*	Progressive weakness of both legs and urinary incontinence for several months	S2/Dura	MSA	Surgery	GR
Mitha *et al.* (2006)[18]	M/42	A 7-year history of low back pain occasionally accompanied by a burning sensation on the back of his left thigh, urinary incontinence	S1/FTI	AFT - ASA (Lt. T9)	Surgery	GR
Witiw *et al.*(2011)[30]	M/62	A 6-month history of progressive bilateral lower extremities paresthesia and weakness, bladder incontinence, and sexual dysfunction	S2-3/FTI	AFT - ASA (Rt. T8)	Surgery	IR
Wajima *et al.* (2014)[29]	M/78	A predominantly progressive numbness of both legs and urinary retention	S1/Dura	AFT-ASA (Rt. T9), Lt. LSA	Embolization with NBCA via both ASA and LSA	GR
Li *et al.* (2017)[16]	M/65**	A 4-year history of progressive weakness and numbness of both legs, erectile dysfunction, BBD	S3/FTI S4/Dura	Upper-AFT-ASA (Lt. L2) Lower-Rt. LSA	Surgery	GR
Iampreechakul *et al.* (2020)[14]	F/52	A 1-year history of LBP with bilateral sciatica, intermittent claudication A 10-month history of progressive paresthesia and weakness of the lower extremities, BBD	S2-3/FTE	LSA (both)	Embolization with NBCA Surgical resection of the filum and intradural draining vein	GR
Ryu *et al.* (2020)[23]	M/68	Progressive myelopathy, BBD	S2-3/FTE	Lt. LSA	Embolization with NBCA	Gradually improved
Present case	M/64	A 3-year history of LBP radiating to both legs, progressive weakness of lower extremities for 3 months, BBD	S2/Dura	LSA (both), MSA	Surgery	GR

AFT-ASA: The artery of the filum terminale arising from anterior spinal artery, BBD: Bowel and bladder dysfunction, F: Female, FTE: Filum terminale externa, FTI: Filum terminale internum, GR: Good recovery, IR: Incomplete recovery, LBP: Low back pain, LSA: Lateral sacral artery, Lt: Left, M: Male, MSA: Middle sacral artery, NBCA: N-butyl cyanoacrylate, Rt: Right, S: Sacral, T: Thoracic, *: Coexisting with the conus AVM, **: Coexisting with the SDAVF at L2 level

The pathogenesis of FTAVF at the sacral region

The exact mechanism of formation of FTAVFs remains controversial. Scullen *et al.*[24] proposed that chronic inflammation, the development of adhesions, and repetitive microtrauma caused by long-standing severe lumbar stenosis may result in hyalinization with fibrosis, limited physiological motion, and increased tension within the FT, inducing the fistula formation. Based on the literature review of 20 cases with FTAVFs in association with spinal canal stenosis by Iampreechakul *et al.*,[14] they found that the level of fistulas in most patients was correlated with the level of spinal stenosis. In addition, Iampreechakul *et al.*[9] described a case of FTAVF at L5 level in association with a large L2-L3 disc sequestration and diffuse lumbar arachnoiditis. They speculated that the formation of this FTAVF may result from severe spinal canal stenosis caused by a large disc sequestration blocking the rostral venous drainage of the fistula, or chronic inflammation, and adhesions of the caudal nerve roots from lumbar arachnoiditis. Based on intraoperative findings, few studies showed the evidence of thickened and opaque arachnoid membrane, adhesions between the nerve roots, and/or chronic inflammation, probably being the precipitating factors involving in the formation of FTAVF.[13,15] In our case, we found the evidence of thickened and opaque arachnoid membrane and adhesions between the cauda equina roots during surgery.

Since 2002, Hsu *et al.*[8] reported a case of concomitant conus medullaris AVM and sacral DAVF of the FT, fed by a dural supply from the MSA. The conus AVM is supplied by the ASA

with venous drainage down to the filum. They suspected that hemodynamic change of spinal cord caused by the spinal cord AVM may produce DAVF of the FT at the level of S2. Similarly, Li *et al*.[16] also reported concomitant SDAVF at the level of L2 and FTAVFs at the level of S3 and S4. In literature review of the coexistence of different spinal vascular malformations in the same patient, venous hypertension and/or thrombosis due to the first spinal lesion at upper lumbar region may produce a second sacral DAVF.[10] Ryu *et al*.[23] reported a case of an AVF located in the FTE at the level of S2-3. They suspected that prolong inflammation and long-standing compression resulting from bilateral sacral perineural cysts may have contributed to the formation of FTE-AVF. In the present study, we speculated that the formation of fistula may cause by venous hypertension secondary to partial thrombosis within the filum vein, probably resulting from long-standing spinal canal stenosis. Imaging studies in our case clearly demonstrated the evidence of partially thrombosis within the dilated vein of the FT rostral to the fistulous point.

The relationship of the fatty FT and FTAVF

FT derived from involution of caudal neural tube. Histologic components of the normal filum may include fibroconnective tissue, neuroglial tissue, peripheral nerve twigs, and adipose tissue.[1] A fatty FT have been occurred due to congenital error in the canalization of the caudal bud of the spinal cord.[20] Tubbs *et al*.[28] demonstrated that mature lobular adipose tissue was present adjacent to the FTE in all cadaveric cases and occasionally it was seen within the FTE. Without the tethered cord syndrome, the presence of adipose tissue in the FT was detected in 17% of the autopsy series.[17]

Since 1989, Djindjian *et al*.[3] reported a middle-aged man with lipoma of the FT and sacral DAVF of the filum at dural sac termination. They speculated that the sacral lipoma may produce local hypervascularization of the dura mater inducing the formation of the acquired fistula. Takai *et al*.[26] presented the radiographic evidence of formation of the FTAVF in a elderly man with a sacral terminal lipoma and tethered cord. During period of 6 years, MRI of the lumbosacral spine revealed the enlargement of lipoma, increased vascular flow voids, and venous congestion of the spinal cord. They speculated that the enlargement of lipoma and increased blood flow may be acquired factors, activating the forming of the fistula located in the FTE. Iampreechakul *et al*.[12] reported an extremely rare case of sacral angiolipoma associated with tethered cord and sacral spina bifida coexisting with sacral AVF draining into the medullary veins through dilated vein of the filum. The enlargement of non-infiltrating angiolipoma may induce thrombosis or impair the venous drainage, activating the formation of the fistula.

In the present study, the findings of transected FT were consistent with the fatty FT. The association between the fatty filum terminale and fistulous formation is not well understood. Recently, Shimizu *et al*.[25] reported two cases of FTAVFs that arose in the fatty FT. Without radiologically evident on preoperative MRI, histological study clearly revealed the adipose tissue around the shunt vessels. They proposed that the lipoma or fatty infiltration of FT may be a predisposing factor for the occurrence of FTAVF, and spinal canal stenosis may secondarily trigger the actual development of the fistula.

Management of sacral DAVF of the FT

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. The key to complete occlusion is obliteration of the proximal vein.[5] Surgical treatment has been the preferred method of treatment with higher complete obliteration rates. Furthermore, surgical obliteration of FTAVFs is technically simple and very effective with low rate of recurrence.[14]

To avoid a longer and more invasive posterior sacrectomy, FTAVF at the sacral region may be treated indirectly by transection of the FT and vessels at lumbar segment rostral to the fistula.[16,18,30] Similarly, our case was indirectly treated by transection of the filum terminale and the draining vein at the level of L5 instead of the level of S2.

CONCLUSION

Sacral DAVFs of the FT are extremely rare. In our case, the formation of fistula may cause by venous hypertension secondary to partial thrombosis within the filum vein, probably resulting from long-standing spinal canal stenosis. Sacral FTAVFs may be found on the pia surface of the terminal FTI, dural component at the area of dural sac termination, or dural extension covering the FTE.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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