Spinal dural arteriovenous fistula associated with L-4 isthmic spondylolisthesis

Case report

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The authors describe a case of a 79-year-old man with a lumbar spinal dural arteriovenous fistula (DAVF) and isthmic spondylolisthesis at the same level. The patient’s thoracic spine MRI study demonstrated swelling and increased T2 signal in the spinal cord and regional dilated perimedullary vessels. Lumbar spine MRI showed L-4 isthmic spondylolisthesis with severe bilateral L4–5 foraminal stenoses. Spinal angiography revealed a fistulous connection at the left L-4 nerve root sleeve between perimedullary veins and a dural branch of the L-4 radicular artery. Based on previous reports about secondary spinal DAVFs, the abnormal vascular communication likely developed secondary to the microtrauma and inflammation on the left L-4 nerve root sleeve, which was attributable to the isthmic spondylolisthesis. The authors performed disconnection of the arteriovenous shunt as well as an L4–5 decompression and posterior instrumented fusion with pedicle screws. The patient’s postoperative course was uneventful, and he improved neurologically. It is important to bear in mind that a spinal DAVF may develop as a consequence of any sort of trauma or inflammation involving nerve roots. One should consider the concomitant treatment of both the spinal DAVF and the underlying pathology that may have given rise to the spinal DAVF.

Key Words • spinal dural arteriovenous fistula • isthmic spondylolisthesis • foraminal stenosis • spinal instability • inflammation • lumbar

Spinal dural arteriovenous fistulas (DAVFs) are the most common type of vascular malformations found within the spinal canal. These lesions are defined as abnormal arteriovenous shunts within the dura that are supplied by the dural arteries and drained by the perimedullary veins.9,14,15 A spinal DAVF is an acquired and progressive condition that typically occurs at very specific locations, such as nerve root sleeves.7 Their etiology remains a matter of debate;7 however, several cases of spinal DAVFs have been reported secondary to previous surgery or trauma.1,2,16,17 We present an unusual case of a spinal DAVF that was fed by the dural branch of an L-4 radicular artery and was associated with L-4 isthmic spondylolisthesis. To our knowledge, a case of spinal DAVF with isthmic spondylolisthesis at the same level has not been previously reported. We discuss the potential association of isthmic spondylolisthesis with the onset of a spinal DAVF and surgical strategy for 2 potentially related but different pathologies.

Case Report

History and Examination. This 79-year-old man without significant medical comorbidities had suffered from constant low-back pain for approximately 40 years following 2 back injuries. His low-back pain was associated with left leg numbness, and his condition had been deteriorating over 3 years. There was also a gradual onset of progressive bilateral leg weakness, gait disturbance, difficulty in balancing, and bladder dysfunction. He consulted an orthopedic surgeon for his back pain and was referred to the neurosurgery service with a presumptive diagnosis of spinal DAVF based on MRI findings.

On examination, there was midline lumbosacral tenderness. The patient reported severe low-back pain combined with left leg numbness. We observed Grade 4 weakness in both legs, more marked on the left, predominantly affecting the tibialis anterior, hamstrings, and quadriceps femoris muscles. The deep tendon reflex was diminished...
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in the quadriceps muscle on the left side. Superficial sensation, temperature, and light touch were decreased in the left L-4 dermatomal distribution and in the sacral levels covering the pudendal and the perianal areas. The patient was not able to maintain an upright position without assistance and was basically confined to a wheelchair because of significantly impaired proprioception and leg weakness on both sides.

Thoracic spine MRI demonstrated swelling with increased T2 signal in the spinal cord from the conus medullaris extending up to T-8 as well as regional dilated perimedullary vessels (Fig. 1A). Lumbar spine MRI showed severe lumbar canal stenosis and an L-4 isthmic spondylolisthesis with bilateral severe foraminal stenoses (Fig. 1B and C). There were fluid collections at the site of the pars defect (Fig. 1C arrow) and Modic Type 1 changes at the L-4 and L-5 vertebral bodies (arrowheads, B and C), indicating significant spinal instability at the L4–5 level. Lumbar spine CT demonstrated bilateral L-4 spondylosis and L4–5 foraminal stenosis with Grade 2 anterolisthesis of L-4 on L-5 (Fig. 1D). Spinal angiography revealed a fistulous connection at the left L-4 nerve root sleeve between the perimedullary veins and the dural branch of the radicular artery arising from the left L-4 segmental artery (Fig. 2). The expanded intradural medullary veins ascended to the level of the conus medullaris, and early venous filling ascended to the level of T-8. We concluded that both mechanical compression associated with foraminal stenosis and lumbar instability at L4–5 contributed to symptomatic development of the spinal DAVF. In this situation, a less invasive simple hemilaminectomy to target just the spinal DAVF was deemed to increase the chance of worsening the patient’s lumbar instability, leading to aggravation of his low-back and leg symptoms. Furthermore, the presence of severe canal stenosis discouraged us from performing a minimal hemilaminectomy for dural opening. After careful consideration, we decided to conduct disconnection of the arteriovenous shunt at the left L-4 nerve root radicular sleeve and perform L4–5 posterior decompression and instrumented fusion with pedicle screws.

**Operation.** We placed pedicle screws into the L-4 and L-5 pedicles bilaterally under Stryker neuronavigation guidance (Stryker Navigation). These screws were all placed with excellent unicortical purchase. We then completed a laminectomy at L-4 and L-5 with a left-sided L4–5 foraminotomy by removing the scar tissue from around the pars defect. We found the L-4 nerve root to be severely compressed at the foramen. The L-4 nerve

![Preoperative images. A: Thoracic spine T2-weighted MR image demonstrating swelling and increased T2 signal in the spinal cord from the conus medullaris extending up to T-8 as well as regional dilated perimedullary vessels. B–D: Lumbar spine T2-weighted MR images (B and C) and CT scan (D) showing L-4 isthmic spondylolisthesis (arrow, D). Severe canal stenosis is seen on the midline section (B). Foraminal stenosis at L4–5 on the left side is obviously visualized on the left parasagittal section (C). There were fluid collections at the site of the pars defect (arrow, C) and Modic Type 1 changes on the L-4 and L-5 vertebral bodies (arrowheads, B and C), indicating significant spinal instability at the L4–5 level.](image)
root was isolated from surrounding scar tissue, followed by coagulation of vessels around the engorged nerve root. The dura was very soft and redundant after the release of chronic compression (Fig. 3A) and was then opened in the midline. The tangled cauda equina was identified, and we proceeded to visualize the affected left L-4 nerve root intradurally and followed it out its foramen. A large distended and tortuous arterialized medullary vein (arrow) is seen at the exit of the L-4 neural foramen. A: The tangled cauda equina was identified after opening the dura. B: A large distended and tortuous arterialized medullary vein (arrow) is seen at the exit of the L-4 neural foramen. C: We placed 2 hemoclips across the vein at the entrance into the dural sleeve, and we cauterized and divided the vein at the point between the 2 clips.

Fig. 3. Intraoperative photographs revealing the dura was very soft and redundant after the release of chronic compression. A: The tangled cauda equina was identified after opening the dura. B: A large distended and tortuous arterialized medullary vein (arrow) is seen at the exit of the L-4 neural foramen. C: We placed 2 hemoclips across the vein at the entrance into the dural sleeve, and we cauterized and divided the vein at the point between the 2 clips.

Postoperative Course. Postoperatively, all neurological symptoms gradually improved over weeks. Postoperative angiography, performed 1 week after surgery, confirmed the disappearance of the spinal DAVF (Fig. 4A). At 8 months after surgery, the patient had continued and significant improvement in all of his symptoms. His back pain and left leg numbness improved dramatically. He was left with some leg weakness but was able to walk independently with a cane. Magnetic resonance images obtained at the 8-month follow-up revealed a marked decrease in the T2 signal in the conus (Fig. 4B), and lumbar spine radiography showed excellent alignment with well-positioned screws (Fig. 4C).

Discussion

This is the first report of a spinal DAVF accompanied by isthmic spondylolisthesis at the same level. Surgical treatment for spinal DAVFs typically requires hemilaminectomy with foraminotomy for intradural disconnection of the AVF; however, the instability of isthmic spondylolisthesis and the presence of severe canal stenosis prompted us to perform instrumented fusion with laminectomy along with occlusion of the fistula.

The unique phenomenon in our case was the association of isthmic spondylolisthesis and the development of a spinal DAVF. The details in this case do not fit the typical demographic pattern of spinal DAVF, which is usually found in the middle thoracic to upper lumbar region. Theoretically, the simultaneous occurrence of the spinal DAVF and isthmic spondylolisthesis may be coincidental; however, we are highly suspicious of a pathophysiological link. Although spinal DAVFs are acquired and progres-
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Most cases are reportedly idiopathic and are independent of the preceding hematological and immunological impairments. However, some pathophysiological mechanisms, such as inflammation, infection, trauma, and thrombosis, are well illustrated as a cause of intracranial DAVFs, which have been more thoroughly studied in previous reports. Several cases of spinal DAVFs have been reported to be secondary to other pathologies (Table 1). These reports speculate that inflammation from direct nerve root injury or impairment of venous return contributed to spinal DAVF formation. Two of 4 patients were in their 20s, which is unusual for idiopathic spinal DAVFs. Asakuno et al. reported only 1 case that was treated surgically. In this case the authors found abnormally massive scar tissue stuck to the nerve root, suggestive of previous nerve root injury. In the present case, we similarly found the nerve root to be very swollen and engorged more than expected in a typical case of isthmic spondylolisthesis.

Some important topics concerning development of spinal DAVFs have been discussed from an anatomical and pathological point of view, which are highly suggestive of the existence of secondary spinal DAVFs. First, Manelfe et al. described normally existing glomerulus-like arteriovenous connections (physiological arteriovenous shunt) predominantly in the thoracolumbar region and located at the dorsal nerve root sleeves between the two layers of dura mater and fed by dural arteries. Second, the valves on the medullary veins, which in normal conditions prevent reflux of blood from extrathecal veins to the coronal venous plexus, are shown to become incompetent under high pressure. Third, in most cases of spinal DAVFs, inflammation is assumed to develop undetected or as an autoimmune reaction. Local inflammation may expand with expression of various cytokines, cause vessel dilation, and open the physiological arteriovenous shunt. This could lead to activation of physiological arteriovenous shunting, resulting in newly formed spinal DAVFs. Based on these facts, we hypothesize that, in the present case, the abnormal vascular communication developed secondary to the inflammation triggered by microtrauma from isthmic spondylolisthesis.

There is some evidence for the existence of inflammation in pseudarthrotic pars defects in patients with lumbar spondylolysis, which can be visualized as fluid collections based on MRI as seen in the present case (Fig. 1C arrow). In addition, Modic Type 1 changes like in the present case, which are hypointense on T1-weighted imaging and hyperintense on T2-weighted imaging, are shown to represent bone marrow edema and inflammation associated with segmental instability. The present case also demonstrated severe long-standing low-back pain and newly developed radiculopathy followed by myelopathy signs. This clinical history is highly suggestive of the direct connection between radicular symptoms caused by long-term spinal instability and development of myelopathy. Constant or intermittent nerve root stretch and impingement as a result of severe foraminal stenosis with instability would add inflammation to the nerve roots. Histological examination of tissues from spondylolysis

Table 1: Reported cases of secondary spinal DAVF

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Level</th>
<th>Age (yrs)</th>
<th>Suspected Cause</th>
<th>Proposed Mechanism</th>
<th>Latency</th>
<th>Proposed Treatment</th>
<th>Surgical Disconnection</th>
<th>Outcome</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yoshino et al., 1998</td>
<td>L-5</td>
<td>27 M</td>
<td>previous L-4-5 anterior interbody fusion</td>
<td>Impaired venous return</td>
<td>7 yrs</td>
<td>Surgical disconnection</td>
<td>Partial resolution</td>
<td>Complete resolution</td>
<td></td>
</tr>
<tr>
<td>Asakuno et al., 2002</td>
<td>L-5</td>
<td>60 M</td>
<td>previous L-5-S1 discoscopy</td>
<td>Nerve root injury</td>
<td>14 mos</td>
<td>Surgical disconnection</td>
<td>Partial resolution</td>
<td>Complete resolution</td>
<td></td>
</tr>
<tr>
<td>Vankan et al., 2004</td>
<td>L-5</td>
<td>60 M</td>
<td>C-1 &amp; C-2 fracture</td>
<td>Nerve root injury</td>
<td>8 mos</td>
<td>Surgical disconnection</td>
<td>Partial resolution</td>
<td>Complete resolution</td>
<td></td>
</tr>
<tr>
<td>Kang et al., 2011</td>
<td>L-4</td>
<td>79 M</td>
<td>L-1 burst fracture</td>
<td>Nerve root injury</td>
<td>Not specified</td>
<td>Surgical disconnection</td>
<td>Partial resolution</td>
<td>Complete resolution</td>
<td></td>
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L-4, 5-isthmic spondylolysis.
defects have shown that the defective ischemic part is well vascularized and rich in perivascularly located nerves.\(^{10}\) This finding may also indicate that angiogenesis is promoted around nerve roots, which could trigger the opening of a dormant physiological arteriovenous shunt at the nerve root sleeve.\(^{10}\)

There are 2 reasons for us to select surgical disconnection of spinal DAVFs. First, endovascular embolization has been generally proposed as an alternative to direct surgery by a number of centers,\(^{11,12}\) but recurrences are not uncommon and more studies and longer-term follow-up are necessary. In addition, adequate coagulation of dural feeders and intradural disconnection of the arterialized vein are all required for complete obliteration and prevention of recurrence,\(^{8}\) which is easily attained by direct surgery. Surgical disconnection is technically simple and very effective.\(^{13,14}\)

Second, the patient in the present case had significant symptomatic instability at the L4–5 level, and inflammation from microtrauma affecting the L-4 nerve root caused by instability was deemed as the main etiology of the spinal DAVF. We judged that the instability required treatment to prevent recurrence of the spinal DAVF. Furthermore, once instrumented fusion was planned, we could perform a wide laminectomy for canal stenosis as well as radical foraminotomy to more easily manipulate the feeding arteries at the nerve root sleeve. Thus, we adopted a combination of surgical disconnection of spinal DAVF and instrumented fusion for L-4 ischemic spondylolisthesis.

It is important to bear in mind the possibility of a spinal DAVF developing as a complication of any sort of trauma or inflammation to nerve roots. Detailed radiographic workup must be performed in cases with progression of neurological deficits that cannot be attributable to the original pathology. One must be certain to treat the newly formed spinal DAVF as well as the condition that caused the DAVF to form in the first place. One should consider a spinal DAVF as a possible etiology when a patient presents with a symptomatic lumbar paraspinal defect and myelopathy signs.

**Disclosure**

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

Author contributions to the study and manuscript preparation include the following. Conception and design: Nishimura, Ginsberg. Acquisition of data: Nishimura. Analysis and interpretation of data: all authors. Drafting the article: Nishimura. Critically revising the article: all authors. Reviewed submitted manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Nishimura. Administrative/technical/material support: Ginsberg. Study supervision: Ginsberg.

**References**


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