Case Report

J Korean Soc Radiol 2021;82(6):1628-1633
https://doi.org/10.3348/jksr.2020.0188
eISSN 2288-2928

대한영상의학회지

Endovascular Treatment of a Lumbar Spinal Epidural Arteriovenous Fistula with Radiculopathy: A Case Report

신경근 압박을 동반한 요추부 척수 경막외 동정맥루의 혈관 내 치료: 증례 보고

Hyun Hwang, MD[1], Jae Ho Shin, MD[2], Jae Taek Hong, MD[3], Yon Kwon Ihn, MD[2]*

[1]Department of Radiology, The Catholic University of Korea, Yeouido St. Mary's Hospital, Seoul, Korea
[2]Department of Radiology, The Catholic University of Korea, St. Vincent's Hospital, Suwon, Korea
[3]Department of Neurosurgery, The Catholic University of Korea, Eunpyeong St.Mary's Hospital, Seoul, Korea

Spinal epidural arteriovenous fistulas (SEDAVFs) are rare spinal vascular malformations that are difficult to diagnose and treat. SEDAVFs can be asymptomatic; however, symptoms can arise from the compression of adjacent nerve roots by dilated vein and perimedullary venous reflux, caused by shunting into the epidural venous plexus. A 31-year-old male presented to our institution with a 2-year history of progressively worsening low-back pain, radiating thigh pain, and sensory changes in his lower extremities. MRI and CT angiography demonstrated dilated epidural vascular lesion compressing the nerve root. The SEDAVF was embolized with multiple coils, which alleviated the nerve root compression from the engorged venous varix and improved the patient's radiculopathy. Our experience from this case shows that endovascular coil embolization using the transarterial approach can be an effective treatment for SEDAVF and an alternative to surgical ligations.

Index terms Fistula; Spine; Arteriovenous Malformations

Received November 7, 2020
Revised February 2, 2021
Accepted March 11, 2021

***Corresponding author**
Yon Kwon Ihn, MD
Department of Radiology,
The Catholic University of Korea,
St. Vincent's Hospital,
93-1 Jungbu-daero, Paldal-gu,
Suwon 16247, Korea.

Tel 82-31-249-7491
Fax 82-31-248-5520
E-mail ihn@catholic.ac.kr

ORCID iDs
Hyun Hwang
https://orcid.org/0000-0003-0564-0990
Jae Ho Shin
https://orcid.org/0000-0001-5922-5720
Jae Taek Hong
https://orcid.org/0000-0001-6453-0439
Yon Kwon Ihn
https://orcid.org/0000-0002-3770-308X

INTRODUCTION

Diagnosis and treatment of spinal epidural arteriovenous fistulas (SEDAVFs) can be difficult because they are rare and diagnosis may require spinal angiography (1-3). SE-

DAVFs can be asymptomatic; however, symptoms can arise from compression of adjacent nerve roots by dilated vein and perimedullary venous reflux, caused by shunting into the epidural venous plexus (1-4). While there are no definite established guidelines for treating SEDAVFs, they may be treated through surgical ligations, endovascular embolization, or a combination of both. In addition to surgical ligations, endovascular embolization via a transarterial or transvenous approach has been shown to be just as effective (2, 4-7). Liquid embolic materials such as glue, onyx, and detachable coils may be utilized for embolization and occlusion of fistula (3, 8, 9).

In this case report, we describe the diagnosis and endovascular coil embolization of a case with a SEDAV in the low lumbar spine, which presented with an engorged venous plexus, leading to severe nerve root compression and radiculopathy.

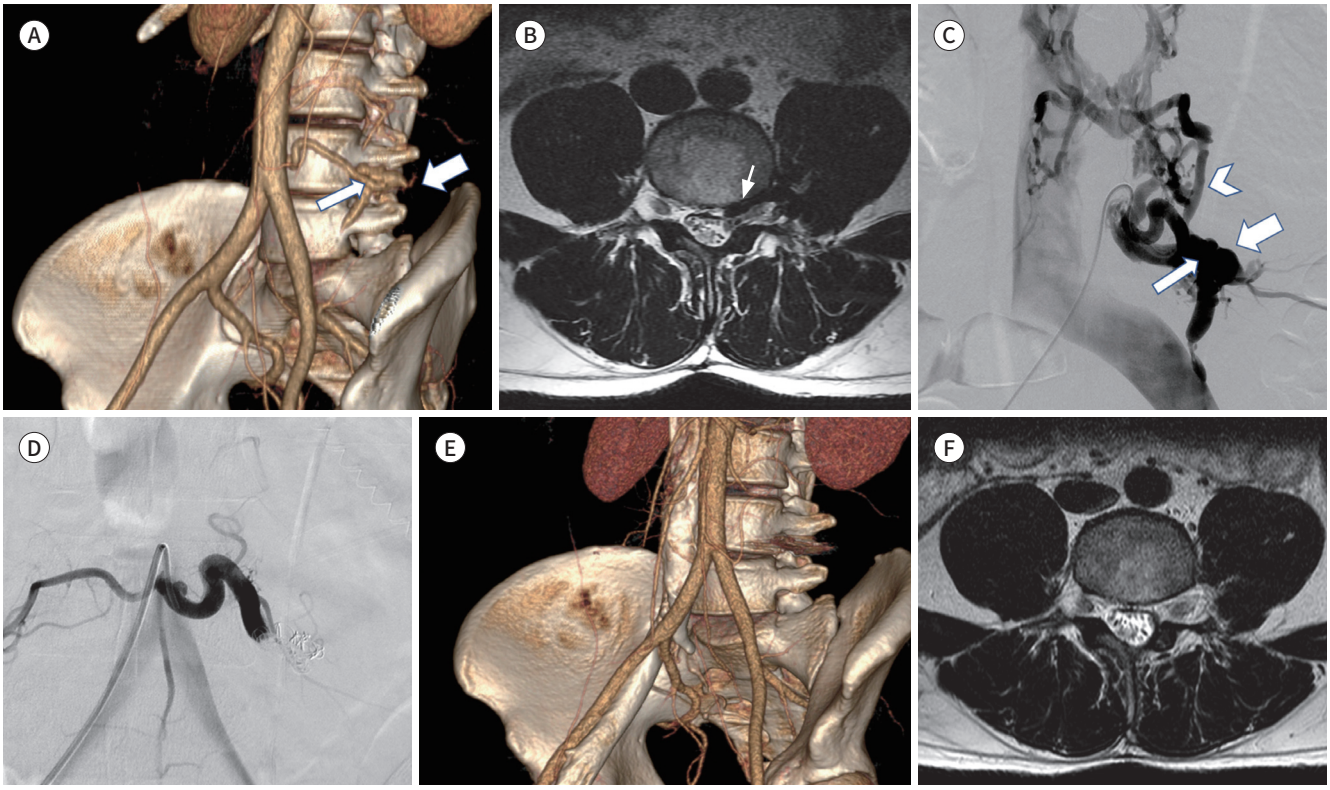
CASE REPORT

This case report was approved by our Institutional Review Board and waived the requirement for informed consent (IRB No. VC19ZESI0214).

A 31–year-old male was presented with worsening low-back pain and radiating thigh pain, which he had been experiencing for two years. He also complained of sensory changes in his left lower extremities. He had no history of trauma or vigorous activities which might have caused the pain. He received nerve block in a local hospital, followed by left partial laminectomy at L4-L5 level due to his low-back pain and spondylodiscitis. Upon admission to our institution, a physical examination indicated a radicular hypoesthesia in the left area at the L4 an L5 level. A plain radiograph showed no remarkable bony or degenerative joint changes. Three-dimensional CT angiography (CTA) demonstrated enlarged lumbar artery and dilated venous pouch in the vicinity of the left L4-L5 foramina (Fig. 1A). T2 weighted MRI scan revealed signal void lesions in the vicinity of the left L4-L5 foramina with dilated epidural vessel which compressed the nerve root (Fig. 1B). No signal changes or enhancement at the spinal cord were observed. Likewise, selective lumbar arteriography showed an enlarged L4 lumbar artery feeding into the shunted pouch (i.e., an arterialized shunting point), which directly drained into engorged, dilated veins and the adjacent epidural venous plexus (Fig. 1C). These radiologic and clinical findings warranted an endovascular approach and embolization of the SEDAVF.

The procedure was conducted under general anesthesia. During the procedure, approximately 4000–5000 IU low molecular weight heparin was utilized. After the procedure, we maintained the same dosage for one day. A 5F guiding catheter (Sheperd hook; Cook, Bloomington, IN, USA) was positioned at the left L4 lumbar artery. Then, selection of the enlarged L4 lumbar artery and selection of proximal site of the arterialized venous shunting point were performed with a microcatheter. (Excelsior SL 10; Stryker, Natick, MA, USA) Using roadmap guidance, a Target 360 coil (7 mm × 30 cm) (Stryker) was deployed at the distal venous portion of the fistula to prevent inadvertent coil migrations. Additional 6 mm × 20 cm and 5 mm × 15 cm coils (Stryker) were sequentially deployed at the draining portion of fistula until the engorged feeding arterial portion of the fistula was sufficiently packed with coils. Sluggish contrast fillings at the fistula and draining venous plexus were observed. After the full de-

Fig. 1. Endovascular coil embolization of a lumbar spinal epidual AVF in a 31-year-old male, presenting with worsening low back pain.
A. 3D CTA of the aorta demonstrates engorged left L4 lumbar artery, shunting point (thin arrow), and abnormally dilated epidural venous pouch (thick arrow).
B. Axial MR T2-weighted image shows prominent abnormal flow void lesions (arrow) occupying the spinal canal and left L4-L5 foramen, compressing the left nerve root.
C. Spinal angiography with anteroposterior projection of left L4 segmental artery shows a spinal epidural AVF without intradural venous drainage. Note the engorged segmental artery, shunting point (thin arrow), dilated epidural venous pouch (thick arrow), draining the ascending lumbar vein (arrowhead), and azygos system (not shown).
D. Final spinal angiography after coil embolization demonstrates total occlusion of AVF and dilated epidural venous pouch.
E. 3D CTA image of the aorta acquired 4 months after the presentation shows complete regression of the engorged left L4 lumbar artery.
F. T2-weighted image demonstrates complete regression of the signal void lesions in the epidural space and preservation of neural foramen at the L4 level.
AVF = arteriovenous fistula, CTA = CT angiography, 3D = three-dimensional



ployment of the coils, a control angiogram demonstrated no evidence that the SEDAVF was opacified (Fig. 1D). After a few days, the patient's symptoms markedly improved and he was discharged. Follow-up CTA, MRI was conducted after 4 months, and it showed that the previously noted enlarged epidural veins and adjacent nerve root compression had resolved (Fig. 1E, F).

DISCUSSION

The exact etiology behind SEDAVF is unclear; however, there is a speculation that spinal surgery may have association with the pathophysiology of SEDAVF such that traumas may cause epidural venous plexus injuries, specifically, microvascular injuries (6, 9). Diagnosing and treating SEDAVFs are challenging because they are rare and their complex pathophysiol-

ogy can be visualized on spinal angiography, and there are no definite established treatment guidelines (3, 8-10). If the SEDVAF drains intradurally, it may cause progressively increasing venous congestion, ischemic myelopathy, and severe back pain (5-7). On the other hand, if SEDAVF drains exclusively epidurally, symptoms may depend on the extent to which the mass impacts the adjacent nerve roots. Recent subcategorization of SEDAVF was proposed Rangel-Castilla et al. (3): SEDAVF with intradural venous drainage (Type A), SEDAVF with exclusive epidural venous drainage with compressive mass-effect (Type B1), and SEDAVF with exclusive epidural venous drainage without compressive mass-effect (Type B2). In our case, radiculopathy was caused by the significant mass-effect from the large arterialized fistula pouch at the epidural venous drainage, compressing the surrounding tissues and adjacent lumbar spine nerve roots (Type B1).

Asymptomatic patients can be closely observed, and there is a very low possibility for spontaneous thrombosis of the draining veins and spontaneous regression of the fistula. Ramanathan et al. (10) reported that one patient out of five patients in their case series demonstrated spontaneous thrombosis of SEDAVF. However, any symptomatic fistula must be treated without delay to prevent perimedullary venous drainage congestion and spinal cord congestion (4-6). Our patient's symptoms resulted from the compression of the nerve root by the arterialized venous varix and therefore warranted urgent treatment by endovascular embolization. The purpose for the endovascular treatment such as coil embolization of SEDAVFs is the occlusion of the fistula between the feeding artery and draining vein (6). In general, the transarterial embolization of the feeding vessel is a minimally invasive and effective method to occlude the fistula when a single arterial feeder is present. Embolization materials such as coils, liquid glues, or onyx can be considered (3, 8, 9). In our case, axial multiplanar reformatted images obtained through angiography clearly showed an exclusive feeding artery for the SEDAVF, which formed a shunt, and compressed the adjacent nerve root. Considering the large feeding artery, the engorged venous pouch, and the large amount of shunt flow, we opted for transarterial approach using detachable coils rather than liquid agents. Some studies have reported liquid agents to be effective for transarterial embolization (3, 8). Although liquid embolic agents may have certain advantages, such as superb penetration of the fistula pouch, there may be a risk for the catheter to be cemented by the cast, potentially causing a vascular injury (3, 8).

Transarterial embolization is preferred over transvenous embolization for accessibility and technical feasibility. As the draining veins of SEDAVFs are located at the azygous venous system, the transvenous approach is technically challenging and often fails due to anatomical complexity. However, transvenous embolization can be appropriate in cases of fistulas with multiple arterial feeders and when there is a risk of injuring the anterior spinal artery (5, 6). Besides the transarterial and transvenous approaches, embolization via direct percutaneous route has also been reported (10). Surgical ligation is generally reserved for difficult, complex lesions where endovascular treatment may result in complications, or for those patients with failed endovascular attempts (5-7, 9, 10). Our case was presented with a large, high flow SEDAVF, which was supplied by a single artery, performing coil embolization with microcatheter possible.

In conclusion, we report a case of SEDAVF which presented with a compressed nerve root.

This case supports that endovascular coil embolization via a transarterial approach is an effective treatment and should be considered as a therapeutic option for SEDAVF.

Author Contributions

Conceptualization, all authors; data curation, all authors; formal analysis, all authors; investigation, all authors; supervision, I.Y.K.; validation, all authors; writing—original draft, all authors; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

Funding

None

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신경근 압박을 동반한 요추부 척수 경막외 동정맥루의 혈관 내 치료: 증례 보고

황 현[1] · 신재호[2] · 홍재택[3] · 인연권[2]*

척수 경막외 동정맥루는 매우 드문 질환으로 진단과 치료가 어렵다. 무증상 환자도 있으나 동정맥단락으로 척수주위정맥으로 역류되고 늘어난 정맥이 인접한 신경근을 압박하면서 증상이 생길 수 있다. 본 증례는 31세 남자 환자로 2년 동안 지속되는 요통, 방사성 허벅지통증, 하지 감각변화로 내원하였다. 컴퓨터단층촬영, 자가공명장치 및 척수혈관조영술에서 요추의 경막외 공간에서 동정맥단락에 의해 늘어난 정맥이 신경근을 압박하고 있는 척수 경막외 동정맥루로 진단하였다. 코일색전술을 시행하여 동정맥단락을 차단하였고 수일 내에 신경근 압박증상이 호전되었다. 이번 증례에서는 척수 경막외 동정맥루에서 외과적 수술의 대안으로 코일 색전술이 효과적인 치료가 될 수 있다는 것을 보여주었다.

[1]가톨릭대학교 여의도성모병원 영상의학과,
[2]가톨릭대학교 성빈센트병원 영상의학과,
[3]가톨릭대학교 은평성모병원 신경외과

https://doi.org/10.3348/jksr.2020.0188