doi: 10.2176/nmccrj.cr.2020-0124

NMC Case Report Journal 8, 229–233, 2021

Spinal Osseous Epidural Arteriovenous Fistula with Intradural Reflux: A Case Report

Kiyoharu SHIMIZU,[1] Takafumi MITSUHARA,[1] Masaaki TAKEDA,[1] Kaoru KURISU,[2] and Satoshi YAMAGUCHI[3]

[1]*Department of Neurosurgery, Graduate School of Biomedical and Health Sciences, Hiroshima University, Hiroshima, Hiroshima, Japan*
[2]*Department of Neurosurgery, Chugoku Rosai Hospital, Hiroshima, Hiroshima, Japan*
[3]*Department of Neurosurgery, University of Iowa Hospitals and Clinics, Iowa City, Iowa, USA*

Abstract

The authors report a rare case of spinal osseous epidural arteriovenous fistula (AVF) with intradural reflux. A 71-year-old lady with a past history of a T12 compression fracture and neurofibromatosis type 1 presented with progressive paraparesis. Magnetic resonance (MR) images of the thoracolumbar spine showed edema of the spinal cord and flow voids. Catheter angiography revealed segmental arteries from T11 to L1 feeding an AVF in the epidural space. The AVF drained not only into the epidural venous plexus but also into the perimedullary veins. Of note, there was an intraosseous drainage route that involved the basivertebral vein of T12. Under the diagnosis of spinal osseous epidural AVF with intradural reflux, surgical interruption of the intradural arterialized draining vein was performed. Spinal osseous epidural AVF with intradural reflux is rare with only four cases reported in the past. We believe that spinal osseous epidural AVF should be recognized as a variant of spinal epidural AVF.

Keywords: spinal osseous epidural arteriovenous fistula, intradural reflux, compression fracture

Introduction

Spinal epidural arteriovenous fistula (AVF) is a rare disease of unknown etiology and pathogenesis.[1)] Spinal arteriovenous shunts are classified based on their vascular supply, drainage, and location of the shunt. Spinal epidural AVFs are classified into two types based on the drainage pattern: the one with intradural reflux and the one with exclusive epidural draiagne.[2,3)] Coexistence of intradural reflux and epidural drainage is very rare.[4)] In addition to this, AVFs that have an intraosseous drainage route are much rarer, with only four cases reported.[5–8)] It has been proposed to distinguish this type of spinal epidural AVF as spinal osseous epidural AVF with intradural reflux.

In this study, we add 5th such case in which spinal osseous epidural AVF with intradural reflux arising in a previous vertebral compression fracture. The characteristics and treatment strategies for spinal osseous epidural AVF with intradural reflux are discussed.

Received April 25, 2020; Accepted September 17, 2020

Case Report

History

A 71-year-old woman presented with a 5-month history of progressive gait disturbance and urinary incontinence. She had a T12 compression fracture about 1.5 years before the manifestation of spinal cord dysfunction. The fracture was managed conservatively, without any neurological consequences. She also had familial neurofibromatosis type 1 that manifested as peripheral nerve tumors in her right thigh and mediastinum. She experienced transient weakness in her legs 4 months before referral to our department. The weakness and dysesthetic pain in her legs became constant and prominent thereafter. When the patient was referred to us, she was unable to walk or even stand without assistance. She also endorsed occasional urinary incontinence. Neurological examination showed grade 3 weakness in the lower extremities with manual muscle testing and decreased sensation below the dermatome of S1.

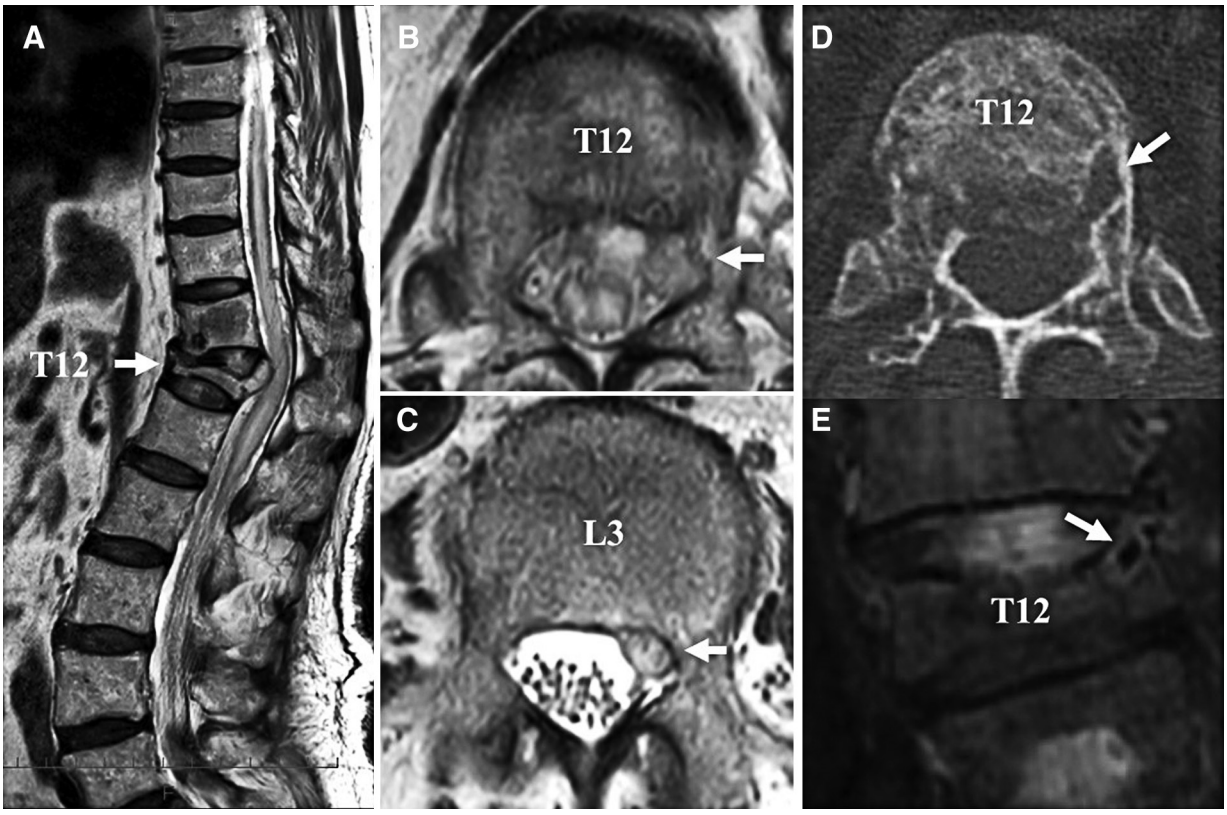

Fig. 1 (A) Sagittal T2-weighted MR image of thoraco-lumbar spine revealed intramedurally high signal intensity of the spinal cord with perimedullary flow voids. (B and C) Axial T2-weighted MR images demonstrated enlarged ventrolateral extradural venous plexus in the spinal canal from T12 to L3 (arrows). (D) Axial CT showed bone defect in the compressed fractured T12 vertebral body (arrow). (E) Coronal fat-suppressed T2-weighted MR image revealed flow voids in the T12 vertebral body in the area consistent with bone defect. CT: computed tomography, MR: magnetic resonance.

Radiological findings

Spinal T2-weighted magnetic resonance (MR) images of the thoracolumbar spine revealed an intramedullary hypersignal intensity area in the spinal cord with perimedullary flow voids (Fig. 1A). Axial T2-weighted MR images demonstrated an enlarged epidural venous plexus ventrolateral to the spinal cord from T12 to L3 (Figs. 1B and 1C). Computed tomography (CT) showed a collapsed T12 vertebral body and a bony defect inside (Fig. 1D). These findings were consistent with those of the chronic phase of a compression fracture. Fat-suppressed T2-weighted MR images revealed flow voids in the bony defect (Fig. 1E). Selective catheter spinal angiography showed epidural AVFs fed by left-sided segmental arteries arising from T11 to L1 (Fig. 2A). The AVFs drained into two pathways: one with intradural reflux via the right L2 radiculomedullary vein and the other into the basivertebral vein of T12 (Figs. 2B and 2C). These findings implied a spinal osseous epidural AVF with intradural reflux.

Treatment and postoperative course

To prevent kyphotic change from worsening, we decided not to perform multilevel laminectomies nor to explore epidural AVFs. Our choice was simple surgical interruption of intradural reflux through a right-sided L1 hemilaminectomy. The arterialized L2 radiculomedullary vein was interrupted with bipolar cauterization. Dysesthetic pain in the lower extremities was improved after the surgery. She became ambulatory with a cane. Postoperative CT angiography confirmed the disappearance of the dilated perimedullary veins (Fig. 3A). MR images obtained 4 months after surgery showed improvement of edema in the spinal cord (Fig. 3B) and shrinkage of the venous pouch in the T12 vertebral body (Figs. 3C and 3D). The kyphotic deformity of the thoracolumbar junction is under close observation.

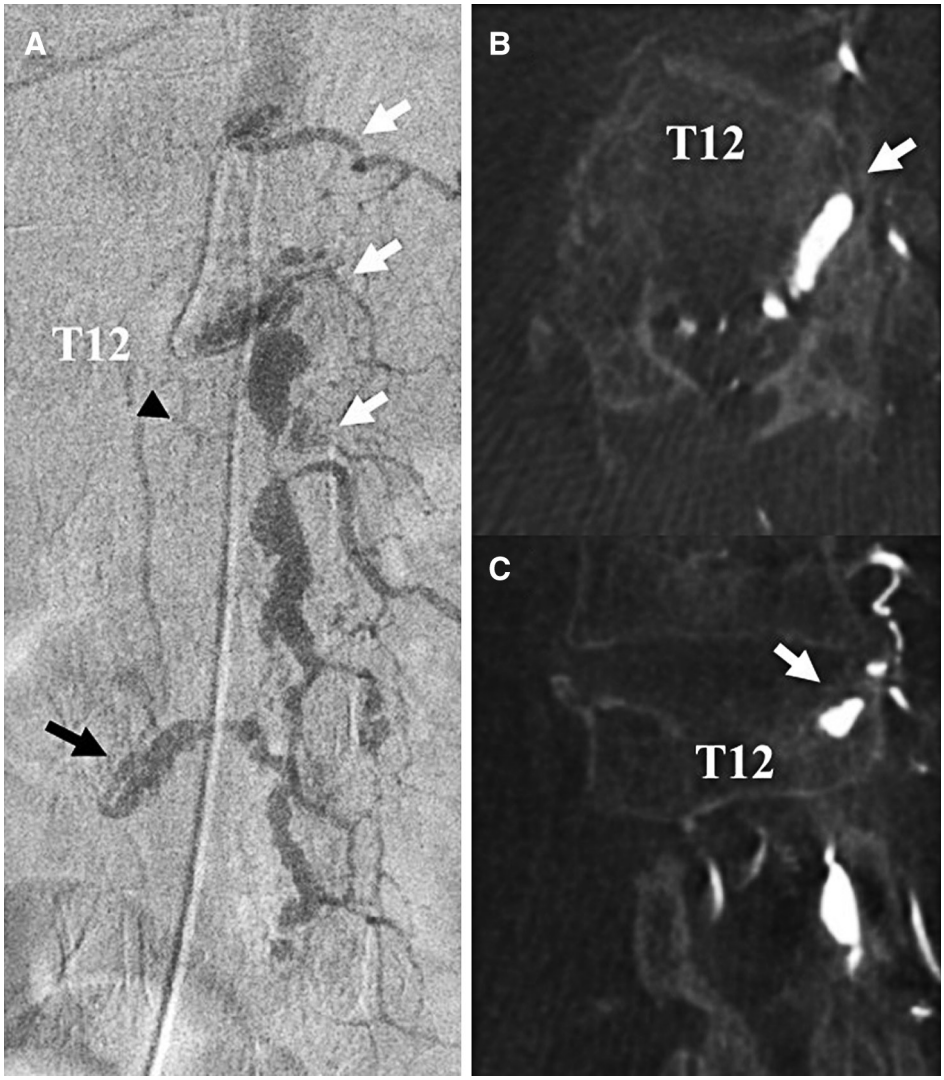

Fig. 2 Selective catheter spinal angiography showed an epidural AVFs fed by the left-sided segmental arteries from T11 to L1 (white arrows) and intradural reflux via the right L2 radiculomedullary vein (black arrow). (A) The spinal epidural AVF had only a single intradural drainer that connecting with perimedullary vein. These findings implied low-flow volume AV shunt (black arrowhead). (B and C) Axial and coronal spinal angiography also showed intraosseous AV fistula connecting to the basivertebral vein of T12 (arrows). AV: arteriovenous, AVFs: arteriovenous fistulae.

Discussion

Spinal osseous epidural AVF with intradural reflux as a variant of epidural AVF

Spinal epidural AVFs have been proposed to be categorized into two types depending on the pattern of venous drainage.[2,3] Type A, that is, spinal epidural AVFs with intradural reflux, is characterized by low-flow volume AV shunt and clinical presentation of congestive myelopathy. Type B, that is, spinal epidural AVF draining exclusively into the epidural venous plexus, is characterized by high-flow volume AV shunt and clinical presentation of compressive myelopathy/radiculopathy by an enlarged epidural venous plexus. Coexistence of intradural reflux and epidural drainage is very rare given the presence of the anti-reflux system of the radicular vein at the dural sleeve and the difference of flow volume between intra- and extradural drainage route.[4,8]

Besides spinal epidural AVFs mentioned above, new entity of spinal osseous epidural AVF in which vertebral body is involved in venous drainage route was first described in 2004.[5–10] Chul et al. distinguished spinal osseous epidural AVF from type B epidural AVF because of the low-flow nature of the AV shunt. In fact, all spinal osseous epidural AVFs

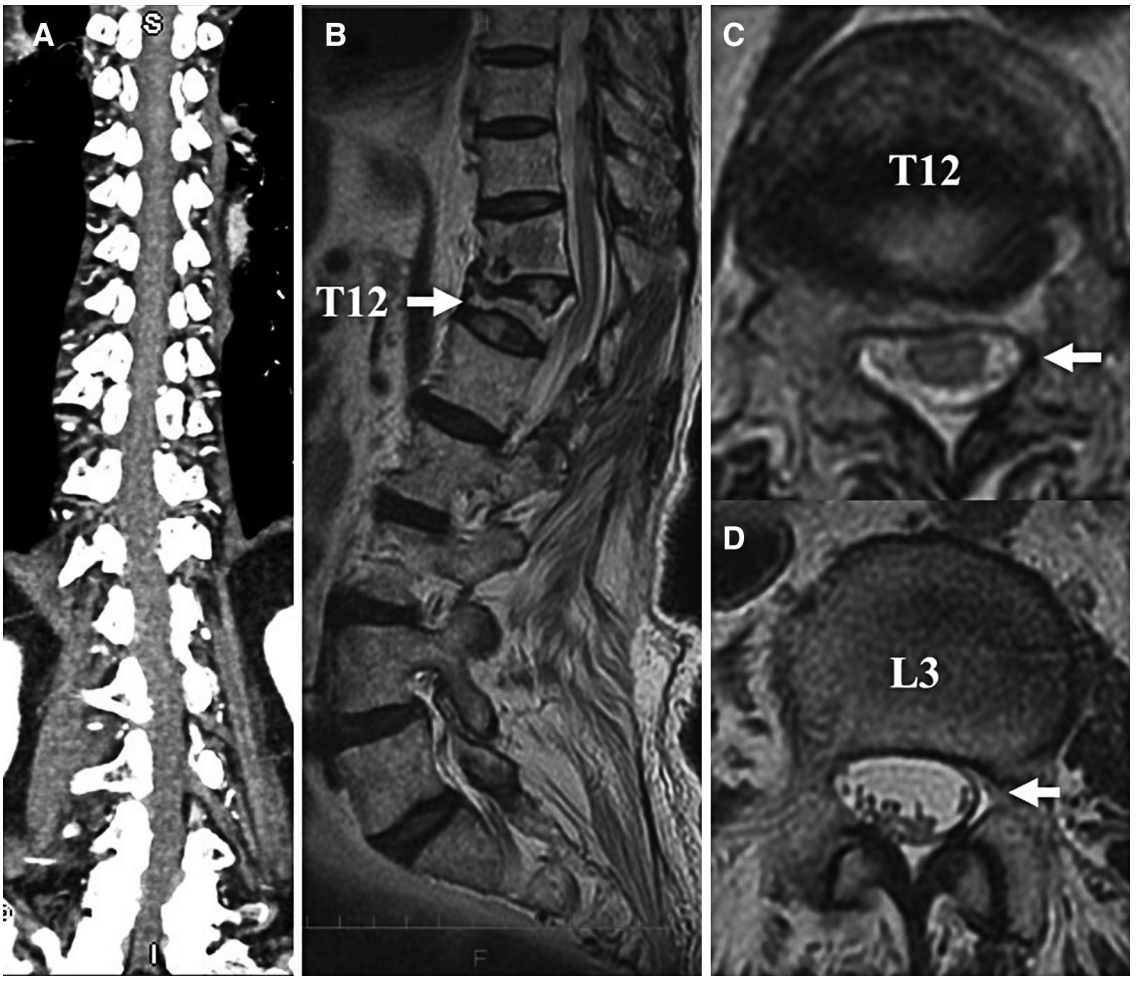

Fig. 3 (A) Coronal CT angiography after the operation revealed the disappearance of the intradural reflux. (B) Sagittal T2-weighted MR image 4 months after the operation showed the attenuation of the high signal intensity in the spinal cord. Kyphotic change at compression fracture was stable. (C and D) Axial T2-weighted MR images revealed the shrinkage of venous pouch (arrow). CT: computed tomography, MR: magnetic resonance.

with intradural reflux had a low-flow AV shunt despite the presence of an enlarged epidural/osseous venous plexus, and they presented with congestive myelopathy. In this regard, spinal osseous epidural AVF with intradural reflux is more similar to type A than to type B epidural AVF. As was assumed by Ou et al., spinal osseous epidural AVF might slowly form inside the fractured vertebral body. It actually took 1.5–9 years for osseous epidural AVFs in previous compression fracture to present with congestive myelopathy.[5–8] This fact tells that osseous epidural AVF could remain asymptomatic for years, until corruption of the reflux-impeding mechanism in the dural sleeve takes place. In fact, two cases of spinal osseous epidural AVF without intradural reflux were found incidentally.[9,10] Both cases were found by screening scan for trauma after motor vehicle accident and femoral neck fracture. These lesions might have been at the stage before corruption of the reflux-impeding mechanism mentioned above. Therefore, spinal osseous epidural AVF, especially without intradural reflux, might be more common and undiagnosed, given that compression fracture is one of the commonest types of spinal trauma.[11,12]

Treatment strategy of spinal osseous epidural AVFs with intradural reflux

Standard treatment of spinal osseous epidural AVFs with intradural reflux is not established yet. Chul et al. reported that transvenous obliteration of AVF with a liquid embolic agent is ideal. However, the venous approach is sometimes impossible when an appropriate venous access route is not found. Transarterial embolization is another option, but is known for a relatively high recurrence rate in the

treatment of spinal epidural AVF.[13] In cases where the venous drainage route extends to the opposite side of the arterial feeders, as in our case, endovascular surgery alone cannot obliterate the shunt. Niizuma et al. discussed the feasibility of successful obliteration of spinal epidural AVF with intradural reflux from the perspective of the number of intradural draining veins. They concluded that simple interruption of intradural reflux could be used in the case of a single intradural draining vein. Occlusion of venous lake, including intradural draining veins, is mandatory in cases with multiple intradural draining veins.[14] Our case had a single intradural venous reflux, and successful obliteration of AVF and intradural reflux was achieved by simple interruption of intradural reflux. However, an accumulation of cases is needed to establish standard treatment for spinal osseous epidural AVF with intradural reflux.

Conclusions

We reported a rare case of spinal osseous epidural AVFs with intradural reflux. Spinal osseous epidural AVFs should be recognized as a variant of spinal extradural AVFs because of their unique angioarchitecture and flow dynamics.

Conflicts of Interest Disclosure

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

References

1) Huang W, Gross BA, Du R: Spinal extradural arteriovenous fistulas: clinical article. *J Neurosurg Spine* 19: 582–590, 2013
2) Rangel-Castilla L, Holman PJ, Krishna C, Trask TW, Klucznik RP, Diaz OM: Spinal extradural arteriovenous fistulas: a clinical and radiological description of different types and their novel treatment with Onyx. *J Neurosurg Spine* 15: 541–549, 2011
3) Takai K, Taniguchi M: Comparative analysis of spinal extradural arteriovenous fistulas with or without intradural venous drainage: a systematic literature review. *Neurosurg Focus* 32: E8, 2012
4) Lenck S, Nicholson P, Tymianski R, et al.: Spinal and paraspinal arteriovenous lesions. *Stroke* 50: 2259–2269, 2019
5) Chul Suh D, Gon Choi C, Bo Sung K, Kim KK, Chul Rhim S: Spinal osseous epidural arteriovenous fistula with multiple small arterial feeders converging to a round fistular nidus as a target of venous approach. *AJNR Am J Neuroradiol* 25: 69–73, 2004
6) Jin YJ, Chung SK, Kwon OK, Kim HJ: Spinal intraosseous arteriovenous fistula in the fractured vertebral body. *AJNR Am J Neuroradiol* 31: 688–690, 2010
7) Gjertsen O, Nakstad PH, Pedersen H, Dahlberg D: Percutaneous intravertebral body embolization of a traumatic spinal epidural arteriovenous fistula with secondary perimedullary venous reflux. A case report. *Interv Neuroradiol* 16: 97–102, 2010
8) Ou CH, Wang HK, Yang TH, Liang CL, Wong HF: Spinal intraosseous epidural arteriovenous fistula with perimedullary drainage obliterated with Onyx embolization: case report. *J Neurosurg Spine* 23: 250–253, 2015
9) Iwakura T, Takehara Y, Yamashita S, et al.: A case of paraspinal arteriovenous fistula in the lumbar spinal body assessed with time resolved three-dimensional phase contrast MRI. *J Magn Reson Imaging* 36: 1231–1233, 2012
10) Imajo Y, Kanchiku T, Yoshida Y, Nishida N, Taguchi T: Large spinal intraosseous arteriovenous fistula: case report. *J Neurosurg Spine* 22: 406–408, 2015
11) Jellema K, Tijssen CC, Sluzewski M, van Asbeck FW, Koudstaal PJ, van Gijn J: Spinal dural arteriovenous fistulas—an underdiagnosed disease. A review of patients admitted to the spinal unit of a rehabilitation center. *J Neurol* 253: 159–162, 2006
12) Johnell O, Kanis JA: An estimate of the worldwide prevalence and disability associated with osteoporotic fractures. *Osteoporos Int* 17: 1726–1733, 2006
13) Kiyosue H, Tanoue S, Okahara M, Hori Y, Kashiwagi J, Mori H: Spinal ventral epidural arteriovenous fistulas of the lumbar spine: angioarchitecture and endovascular treatment. *Neuroradiology* 55: 327–336, 2013
14) Niizuma K, Endo T, Sato K, et al.: Surgical treatment of spinal extradural arteriovenous fistula with parenchymal drainage: report on 5 cases. *Neurosurgery* 73: onsE287–283; discussion onsE293-284, 2013

Corresponding author: Kiyoharu Shimizu
Department of Neurosurgery, Graduate School of Biomedical and Health Sciences, Hiroshima University, 1-2-3 Kasumi, Minami-ku, Hiroshima, Hiroshima 734-8551, Japan.
e-mail: shimizu.kiyoharu@gmail.com