



HOKKAIDO UNIVERSITY

Title	Cervical Epidural Arteriovenous Fistula With Radiculopathy Mimicking Cervical Spondylosis : Case Report
Author(s)	Kawabori, Masahito; Hida, Kazutoshi; Yano, Shunsuke et al.
Citation	Neurologia medico-chirurgica, 49(3), 108-113 https://doi.org/10.2176/nmc.49.108
Issue Date	2009-03
Doc URL	https://hdl.handle.net/2115/70764
Rights(URL)	https://creativecommons.org/licenses/by-nc-nd/4.0/
Type	journal article
File Information	E6_49_108.pdf



Cervical Epidural Arteriovenous Fistula With Radiculopathy Mimicking Cervical Spondylosis

—Case Report—

Masahito KAWABORI, Kazutoshi HIDA, Shunsuke YANO,
Takeshi ASANO, and Yoshinobu IWASAKI

*Department of Neurosurgery, Hokkaido University Graduate School of Medicine,
Sapporo, Hokkaido*

Abstract

A 65-year-old woman presented with a rare case of cervical epidural arteriovenous fistula (AVF) manifesting as radiculopathy of the right upper extremity that mimicked cervical spondylosis. She had a 2-month history of gradually progressive right-hand motor weakness and sensory disturbance. The initial diagnosis was cervical disk herniation. However, computed tomography with contrast medium showed abnormal enhancement at the right C5-6 and C6-7 intervertebral foramina. Magnetic resonance (MR) imaging with gadolinium disclosed an enhanced abnormal epidural mass at the dorsal surface of the dural tube between the C5 and C6 vertebrae. T₂-weighted MR imaging showed a slight flow void on the dorsal and ventral surfaces of the spinal cord between C3 and T4. Digital subtraction angiography disclosed cervical epidural and dural AVFs fed by the C5 and C6 radicular arteries. The diagnosis was concomitant epidural and dural AVFs. The dilated internal vertebral venous plexus attributable to epidural AVF was considered to be responsible for the radiculopathy. Transarterial embolization using n-butylcyanoacrylate achieved complete occlusion of the lesions. Her symptoms improved immediately and MR imaging and angiography performed 10 days postembolization showed reduction of both the epidural and dural AVFs.

Key words: cervical arteriovenous fistula, epidural arteriovenous fistula, radiculopathy, endovascular embolization

Introduction

Classification of spinal arteriovenous malformations (AVMs) remains controversial,^{1,9)} but may include 4 types, intramedullary AVM, perimedullary arteriovenous fistula (AVF), dural AVF with intradural drainage, and dural AVF with extradural drainage (so-called epidural AVF).¹⁹⁾ Dural AVF¹⁶⁾ is the most common form of spinal AVM, accounting for 50–85%.^{2,22)} Dural AVF is fed by the radiculomeningeal artery and the venous drainage pattern is exclusively retrograde toward the spinal cord via the perimedullary vein. Dural AVF usually manifests as slowly progressive myelopathy caused by venous hypertension and spinal cord congestion.^{3,9,22)} Epidural AVF accounts for approximately 1.6% of all spinal AVMs,³⁾ and drains mostly into the internal vertebral venous plexus.^{1–6,8–15,17,20–22,24,26,27)} This

type of AVF is also known as extradural, paravertebral, and paraspinal AVF.^{1–6,8–15,17,20,22,24,26,27)} Our series of 137 patients with spinal AVM treated since 1986 includes 22 (16%) with intramedullary AVM, 45 (33%) with perimedullary AVF, 65 (47%) with dural AVM, and 5 (4%) with epidural AVF.

Here we report a rare case of cervical epidural AVF manifesting as radiculopathy of the right upper extremity treated by interventional surgical procedure.

Case Report

A 65-year-old woman experienced gradual worsening of dysesthesia and numbness of the 1st to 3rd fingers of the right hand. One month later she noticed weakness of the right arm and within the next month her condition progressed to monoplegia. A provisional diagnosis of cervical disk herniation was made at a local hospital. Computed tomography (CT) with contrast medium showed abnormal signs, so

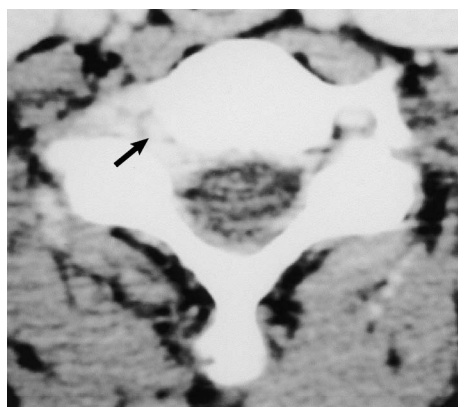


Fig. 1 Axial computed tomography scan with contrast medium showing a well-enhanced lesion at the right C5-6 intervertebral foramen (arrow).



Fig. 2 A: Sagittal T₂-weighted magnetic resonance image revealing multiple flow voids on both the dorsal and ventral surfaces of the spinal cord from the C3 to T4 levels (arrows), and slight swelling of the spinal cord without abnormal signal intensity at the C3-C6 levels. B: Sagittal T₁-weighted magnetic resonance image with gadolinium showing an abnormal enhanced epidural mass located ventral to the dural tube at the C5-C6 levels (arrowheads).

she was referred to our institute.

On admission, neurological examination revealed

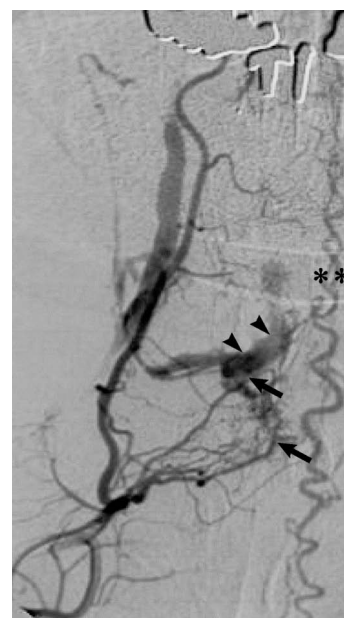


Fig. 3 Digital subtraction angiogram of the right thyrocervical trunk showing filling of the internal vertebral venous plexus indicative of a cervical epidural arteriovenous fistula (arrows) fed by multiple feeders from the C5-C6 radicular arteries, and drained mainly into the internal vertebral venous plexus (arrowheads) and partly into the medullary veins (asterisks) running into the dilated perimedullary vein.

slight motor weakness of the right deltoid, biceps, and brachioradial muscles and hypesthesia in the area of the C5-C6 dermatomes of the right arm. The deep tendon reflexes of the right biceps and brachioradial muscles were diminished. CT with contrast medium showed an irregular enhanced lesion at the right intervertebral foramen of C5-6 and C6-7 with extension into the right ventrolateral epidural space (Fig. 1). Magnetic resonance (MR) imaging revealed multiple flow voids on the surface of the spinal cord from the C3 to T4 levels and slight swelling of the spinal cord without abnormal signal intensity (Fig. 2A). MR imaging with gadolinium showed an enhanced abnormal epidural mass ventral to the dural tube at the C5-C6 levels (Fig. 2B). Selective angiography disclosed a cervical dural AVF fed mainly by the C5-C6 radicular arteries diverging from the right thyrocervical trunk, and drained primarily into the internal vertebral venous plexus and partly into the perimedullary veins. Marked dilation of the internal vertebral venous plexus was observed (Fig. 3). The diagnosis was concomitant epidural and dural AVFs. We thought that

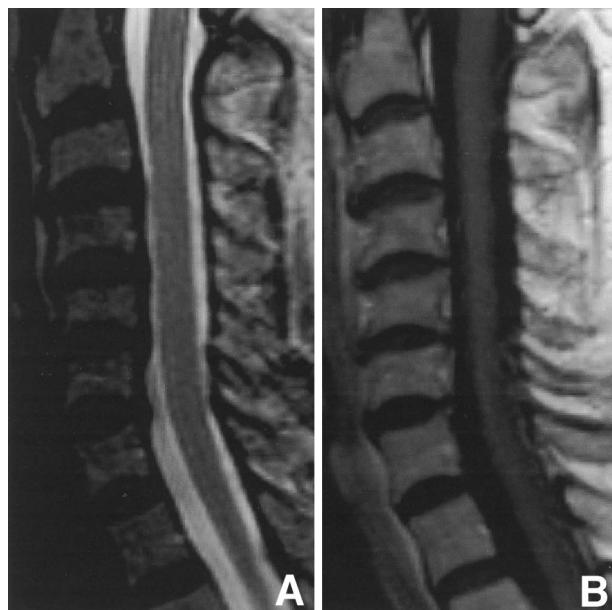


Fig. 4 A: Sagittal T₂-weighted magnetic resonance image showing disappearance of the spinal cord swelling and multiple flow voids on the spinal cord. B: Sagittal T₁-weighted magnetic resonance image with gadolinium showing no abnormal enhancement of the ventral to cervical cord.



Fig. 5 Digital subtraction angiogram of the right thyrocervical trunk obtained 10 days postembolization showing complete disappearance of the fistulae.

the radiculopathy was caused by compression of the nerve root by the dilated epidural venous plexus, so endovascular embolization was planned.

Multiple feeders of the AVF were embolized with a mixture of n-butyl cyanoacrylate and lipiodol using a 1.2-French catheter. The first attempt to obtain complete embolization failed due to the presence of many feeding arteries, but blood flow to the AVF was markedly reduced. Postembolization, her neurological condition was remarkably improved, with decreased right-arm weakness (manual muscle test 4/5) and complete disappearance of the sensory disturbances. T₂-weighted MR imaging obtained 4 days after embolization showed disappearance of the spinal cord swelling and the abnormal enhancement at the ventral side of the spinal cord (Fig. 4). Selective angiography performed 10 days later confirmed complete occlusion of the arteriovenous shunt (Fig. 5). We continue to monitor her condition on an out-patient basis.

Discussion

The clinical characteristics, treatment, and outcome in the 31 reported patients with epidural AVF, including ours, are summarized in Table 1. There are some interesting differences between dural and epidural AVFs. Patients with epidural AVF tend to be younger than patients with dural AVF (mean age 37 vs. 49 years),²³⁾ and 97% of dural AVFs were located in the lower thoracic and lumbar regions,²³⁾ whereas 58% of epidural AVFs occurred in the cervical region.¹⁰⁾ Dural AVF tended to be fed by a single artery with low blood flow through the fistula, whereas epidural AVF was fed by multiple arteries with rapid blood flow.²³⁾ Spinal dural AVF manifested as slowly progressive myelopathy caused by spinal congestion, whereas epidural AVF manifested as myelopathy caused by the mass effect of the dilated internal vertebral venous plexus or radiculopathy caused by compression of the dilated internal vertebral venous plexus or inadequate venous drainage from the root sleeve.^{5,7,9,27)}

The pathogenesis of both dural and epidural AVFs is not yet clearly understood. The AVF may be acquired by the process of recanalization.^{7,18,25)} Dural trauma inflicted by an adjacent intervertebral disk, bone, or ligament results in dural venous thrombosis, and the presence of the dural arterial plexus and rich capillary network near a draining thrombosed vein may lead to recanalization of the venous system via neovascularization, resulting in a shunt system.²²⁾ The presence of epidural venous drainage may be explicable by total recanalization of the epidural system, but this hypothesis fails to explain the differences in the etiology of dural and epidural AVFs, suggesting that some other factors may be involved.

Table 1 Reported cases of epidural arteriovenous fistula

Author (Year)	Age (yrs), Sex	Radicu- lopathy	Mye- lopathy	Site	Feeding arteries	Draining veins	Communica- tion with intradural structures	Treatment	Outcome
Bradac et al. (1977) ⁽⁴⁾	41, F	+	+	C6-T1	VA	IVVP, IMV	+	surgery	good
Deans et al. (1982) ⁽⁸⁾	53, F, NF	-	+	C2-C4	VA	IVVP	-	surgery	good
	45, F, NF	+	+	C4-5	VA	IVVP	-	embo.	good
Han et al. (1987) ⁽¹⁰⁾	27, M	+	-	L3-L4	lumbar A	IVVP	-	embo., surgery	good
Cahan et al. (1987) ⁽³⁾	51, F	+	-	C4-C5	VA, thyro- A, costo- A	IVVP	-	embo.	good
	19, M	-	-	C5	VA	IVVP, IMV	+	embo.	good
Heier and Lee (1987) ⁽¹²⁾	78, M	-	+	L	sacral A	IVVP	-	embo.	good
Johnson et al. (1990) ⁽¹⁵⁾	11, F, NF	-	-	C2-C3	VA	IVVP	-	embo.	good
Willinsky et al. (1990) ⁽²⁶⁾	57, M	+	+	C4-C5	VA, thyro- A, costo- A	IVVP	-	embo.	good
	41, M	-	+	T7-T10	intercostal A	IVVP	-	embo.	good
Hui et al. (1994) ⁽¹³⁾	10, F	-	+	C6-C7	VA	IVVP	+	embo.	moderate improvement
	1, M	-	-	T6-T7	intercostal A	IVVP, intercostal V	-	embo.	good
	1, F	-	-	C	thyro- A	IVVP	-	embo.	good
Ikeda et al. (1994) ⁽¹⁴⁾	62, F	-	-	C2	VA	IVVP, IMV	+	surgery	good
Arnaud et al. (1994) ⁽¹¹⁾	60, M	-	+	T7-T12	lumbar A	IVVP	+	embo.	moderate improvement
	17, F	+	-	L4-L5	iliolumbar A	IVVP	+	embo.	good
Cognard et al. (1995) ⁽⁷⁾	51, M	+	+	C2-C5	VA, thyro- A, costo- A	IVVP	-	embo., surgery	good
Kohno et al. (1996) ⁽¹⁷⁾	72, M	-	+	S1	sacral A	IVVP, IMV	+	embo., surgery	good
Pirouzmand et al. (1997) ⁽²²⁾	73, M	-	+	L	lateral sacral A	IVVP, IMV	+	surgery	good
Goyal et al. (1999) ⁽⁹⁾	68, M	-	+	C	VA, thyro- A, costo- A	IVVP	-	embo.	good
	70, M	-	+	T12	intercostal A	IVVP, IMV	+	surgery	good
	7, M	-	-	T5	intercostal A	IVVP	-	embo.	good
	13, F	-	+	T11-T12	intercostal A	IVVP	-	no treatment	
	17, F	-	+	T9-L1	intercostal A, lumbar A	IVVP	-	embo.	poor
Asai et al. (2001) ⁽²⁾	24, M	-	+	C5-T2	thyro- A, costo- A	IVVP	-	embo., surgery	good
Chuang et al. (2003) ⁽⁶⁾	4, F	-	+	C6-C7	costo- A	IVVP	-	embo., surgery	moderate improvement
	4, F	+	-	C6-T2	intercostal A	IVVP	-	surgery	good
Hauck and Nauta (2006) ⁽¹⁵⁾	51, F, NF	+	+	C4-C7	VA, thyro- A, costo- A	IVVP	-	embo., surgery	good
Saito et al. (2007) ⁽²⁴⁾	62, F, NF	-	+	T10-T11	intercostal A	IVVP, intercostal V	+	surgery	moderate improvement
Paolini et al. (2008) ⁽²¹⁾	26, F, NF	-	+	C2-C3	VA, thyro- A	IVVP	-	embo., surgery	good
Present case	65, F	+	-	C5-C7	thyro- A	IVVP	+	embo.	good

A: artery, costo-: costocervical trunk, embo.: embolization, IMV: intramedullary vein, IVVP: internal vertebral venous plexus, NF: neurofibromatosis, thyro-: thyrocervical trunk, V: vein, VA: vertebral artery, +: present, -: absent.

Some patients with epidural AVF, including ours, present with both epidural and intradural drainage. Epidural flow is frequently rapid, whereas intradural flow is slow and retrograde.²³⁾ This phenomenon may be explained by the existing venous drainage pathways of the cord.⁸⁾ The perimedullary veins draining the spinal cord usually open into the spinal-nerve venous channels at the point where they pierce the dura. The epidural venous system often has segmental communications with spinal-nerve venous channels. The radicular vein forms this type of interconnection with the internal vertebral venous plexus in patients with reflux into the perimedullary vein. In contrast, no drainage into the perimedullary vein occurs despite gross dilation of the epidural veins in patients with radicular veins drained by an alternative route without interconnection with the epidural vein. Intradural communication may develop after prolonged chronic high pressure from the arterial epidural venous plexus.⁹⁾ An anti-reflux mechanism may exist in the radicular veins at the point where they penetrate the dura.^{18,25)} These vessels are thought to function in the maintenance of constant venous pressure in the spinal cord regardless of changes in the intra-abdominal or intra-thoracic pressure. Chronic hypertension in the epidural space may result in failure of this anti-reflux mechanism.

The treatment of asymptomatic epidural AVF with multiple feeders remains challenging. Endovascular rather than open surgery is the initial treatment of choice,²⁶⁾ but embolization of the feeding arteries without complete obliteration of the draining vein may result in further recruitment of feeders.⁹⁾ Use of a venous access route may produce dramatic clinical improvement so this approach should be considered as part of the treatment strategy.²⁷⁾

Spinal AVM is rare and epidural AVF is even more rare, but these lesions should be considered in patients presenting with radiculopathy mimicking cervical spondylosis. Therefore, in the absence of cervical spondylosis, additional examinations such as MR imaging with gadolinium should be performed.

References

- 1) Arnaud O, Bille F, Pouget J, Serratrice G, Salamon G: Epidural arteriovenous fistula with perimedullary venous drainage: case report. *Neuroradiology* 36: 490-491, 1994
- 2) Asai J, Hayashi T, Fujimoto T, Suzuki R: Exclusively epidural arteriovenous fistula in the cervical spine with spinal cord symptoms: case report. *Neurosurgery* 48: 1372-1376, 2001
- 3) Bao YH, Ling F: Classification and therapeutic modalities of spinal vascular malformations in 80 patients. *Neurosurgery* 40: 75-81, 1997
- 4) Bradac GB, Simon RS, Schramm J: Cervical epidural AVM. Report of a case of uncommon location. *Neuroradiology* 14: 97-100, 1977
- 5) Cahan LD, Higashida RT, Halbach VV, Hieshima GB: Variants of radiculomeningeal vascular malformations of the spine. *J Neurosurg* 66: 333-337, 1987
- 6) Chuang NA, Shroff MM, Willinsky RA, Drake JM, Dirks PB, Armstrong DC: Slow-flow spinal epidural AVF with venous ectasias: two pediatric case reports. *AJNR Am J Neuroradiol* 24: 1901-1905, 2003
- 7) Cognard C, Semaan H, Bakchine S, Miaux Y, Thibault S, Sola Martinez MT, Chiras J: Paraspinal arteriovenous fistula with perimedullary venous drainage. *AJNR Am J Neuroradiol* 16: 2044-2048, 1995
- 8) Deans WR, Bloch S, Leibrock L, Berman BM, Skultety FM: Arteriovenous fistula in patients with neurofibromatosis. *Radiology* 144: 103-107, 1982
- 9) Goyal M, Willinsky R, Montanera W, terBrugge K: Paravertebral arteriovenous malformations with epidural drainage: clinical spectrum, imaging features, and results of treatment. *AJNR Am J Neuroradiol* 20: 749-755, 1999
- 10) Han SS, Love MB, Simeone FA: Diagnosis and treatment of a lumbar extradural arteriovenous malformation. *AJNR Am J Neuroradiol* 8: 1129-1130, 1987
- 11) Hauck EF, Nauta HJ: Spontaneous spinal epidural arteriovenous fistulae in neurofibromatosis type-1. *Surg Neurol* 66: 215-221, 2006
- 12) Heier LA, Lee BC: A dural spinal arteriovenous malformation with epidural venous drainage: a case report. *AJNR Am J Neuroradiol* 8: 561-563, 1987
- 13) Hui F, Trossello MP, Meisel HJ, Alvarez H, Sequeira E, Lasjaunias P: Paraspinal arteriovenous shunts in children. *Neuroradiology* 36: 69-73, 1994
- 14) Ikeda H, Fujimoto Y, Koyama T, Fujimoto Y: [A rare case of high cervical spinal cord dural arteriovenous fistula presenting with intracranial subarachnoid hemorrhage]. *No Shinkei Geka* 22: 1045-1048, 1994 (Jpn, with Eng abstract)
- 15) Johnson CE, Russell EJ, Huckman MS: Resolution of spinal epidural vascular pseudotumor following balloon occlusion of a postoperative vertebral arteriovenous fistula. *Neuroradiology* 31: 529-532, 1990
- 16) Kendall BE, Logue V: Spinal epidural angiomatous malformations draining into intrathecal veins. *Neuroradiology* 13: 181-189, 1977
- 17) Kohno M, Takahashi H, Ide K, Ishijima B, Yamada K, Nemoto S: A cervical dural arteriovenous fistula in a patient presenting with radiculopathy. Case report. *J Neurosurg* 84: 119-123, 1996
- 18) Manelfe C, Lazorthes G, Roulleau J: [Arteries of the human spinal dura mater]. *Acta Radiol Diagn (Stockh)* 13: 829-841, 1972 (Fre)
- 19) Miyasaka K, Asano T, Ushikoshi S, Hida K, Koyanagi

- I: Vascular anatomy of the spinal cord and classification of spinal arteriovenous malformations. *Interventional Neuroradiology* 6 (Suppl 1): 195–198, 2000
- 20) Oda Y, Konishi T, Suzui H, Fujimoto N, Nakamura K, Matsumoto M, Okumura T, Kang Y, Kikuchi H: [Partially thrombosed radiculomeningeal arterio-venous fistula in spinomedullary junction]. *No Shinkei Geka* 17: 63–68, 1989 (Jpn, with Eng abstract)
 - 21) Paolini S, Colonnese C, Galasso V, Morace R, Tola S, Esposito V, Cantore G: Extradural arteriovenous fistulas involving the vertebral artery in neurofibromatosis Type 1. *J Neurosurg Spine* 8: 181–185, 2008
 - 22) Pirouzmand F, Wallace MC, Willinsky R: Spinal epidural arteriovenous fistula with intramedullary reflux. Case report. *J Neurosurg* 87: 633–635, 1997
 - 23) 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* 67: 795–802, 1987
 - 24) Saito A, Takahashi T, Ezura M, Tominaga T: Inter-costal arteriovenous fistula associated with neurofibromatosis manifesting as congestive myelopathy: case report. *Neurosurgery* 61: E656–E657, 2007
 - 25) Tadie M, Hemet J, Aaron C: Le dispositif protecteur antireflux des veins de la moelle. *Neurochirurgie* 25: 28–30, 1979 (Fre)
 - 26) Willinsky R, TerBrugge K, Lasjaunias P, Montanera W: The variable presentations of craniocervical and cervical dural arteriovenous malformations. *Surg Neurol* 34: 118–123, 1990
 - 27) Willinsky R, terBrugge K, Montanera W, Wallace MC, Gentili F: Spinal epidural arteriovenous fistulas: arterial and venous approaches to embolization. *AJNR Am J Neuroradiol* 14: 812–817, 1993

Address reprint requests to: Masahito Kawabori, M.D.,
Department of Neurosurgery, Hokkaido University
Graduate School of Medicine, North-15 West-7,
Kita-ku, Sapporo 060-8638, Japan.
e-mail: masahitokawabori@yahoo.co.jp