N-Butyl 2-Cyanoacrylate Embolization of Spinal Dural Arteriovenous Fistula Presenting with Subarachnoid Haemorrhage

B.J. KWON, T.-K. KIM*, S.I. SEO*, J.B. KYUNG*, H.Y. SEOL*, M.H. HAN

Department of Radiology, Seoul National University College of Medicine, Seoul; Korea

**Department of Diagnostic Radiology, Korea University, College of Medicine, Seoul; Korea*

Key words: spinal dural arteriovenous fistulae; N-butyl 2-cyanoacrylate embolization; subarachnoid haemorrhage

Summary

We report an unusual case of spinal dural arteriovenous fistula (SDAVF) presenting with subarachnoid haemorrhage (SAH). Cure was achieved with endovascular treatment with n-butyl 2-cyanoacrylate (NBCA). A review of the literature revealed five cases of cervical SDAVF that presented with SAH. None of these cases were treated with NBCA.

Introduction

Spinal dural arteriovenous fistulae (SDAVF) are usually located in the thoracolumbar region¹. SDAVF in the cervical spine is rare, and when present usually occurs with myelopathy², which is much the same as SDAVF in the thoracolumbar spine. However, some of the reported cases of cervical SDAVF have presented with subarachnoid haemorrhage (SAH). We found five cases of cervical SDAVF presenting with SAH by literature review³⁻⁷. Cure was achieved by surgical ligation in four cases and by using a single detachable silicone balloon in one case. We report an unusual case of SDAVF in the cervical spine accompanied by SAH that was cured successfully by endovascular treatment with n-butyl 2-cyanoacrylate (NBCA).

Case Report

A 32-year-old man with no pertinent medical history came to the emergency department with

a sudden loss of consciousness. He awoke the next morning with severe nuchal rigidity, headache and nausea. A CT scan of the head revealed SAH in the basal cisterns, sylvian fissures, and prepontine cistern (figure 1A). He was neurologically intact. Angiography showed high-flow vertebral arteriovenous fistulae at the left C5-C6 intervertebral level. The nidus of the lesion was supplied by radicular branches of the vertebral and deep cervical arteries, which shunted into a varix adjacent to a nerve root. Venous drainage was via the coronal venous system on the dorsal surface of the spinal cord. The enlarged coronal venous system flowed in both upward and downward directions. MR images of the cervical, thoracic, and lumbar spine showed enlarged serpentine vascular structures on the dorsal surface of the cord from the cervicomedullary junction to the T3 level. No abnormality was evident within the cord (figure 1).

Interventional neuroradiological treatment was initiated by superselective catheterization using a microcatheter with the aid of digital fluoroscopy and roadmapping. Digital subtraction angiography runs were performed to improve visualization of the fistula and to exclude the presence of anterior or posterior spinal arteries. When the microcatheter was positioned as closely to the fistula as possible, multiple test contrast material injections were done, and then, using a single-column technique, 0.2 to 0.7 cc of a 33% diluted NBCA: ethiodol mixture was injected under direct fluoroscopic guid-

Figure 1 Pre-treatment study of a 32-year-old man with loss of consciousness, nuchal rigidity and severe headache. A) CT scan shows subarachnoid haemorrhage in the basal cistern. A) anteroposterior (B) and lateral (C) angiograms of the left vertebral artery reveal a spinal dural arteriovenous fistula fed by the radicular artery and multiple muscular branches at the C5-C6 intervertebral level. Note the retrograde filling of deep cervical arteries. The draining vein forms a varix adjacent to a nerve root. The venous drainage was via the coronal venous system on the dorsal surface of the spinal cord. D) T2-weighted MRI scan shows numerous signal voids on the dorsal surface of the cervical cord. No signal change was observed within the cord.

ance. We also used fiber coils, if embolization with liquid material was deemed unsafe, depending on the microcatheter position. Embolization was ended when the NBCA: ethiodol mixture was clearly documented to be in the fistula and the variceal draining vein. After embolization, a repeat angiogram revealed no residual fistula (figure 2).

Postinterventional T1-weighted MR images showed high signal intensities, indicating the NBCA: ethiodol mixture within the thrombosed veins instead of serpentine vascular signal voids on the dorsal surface of the cervical cord (figure 2C). The patient was asymptomatic after the procedure and no residual fistula was identified on the follow-up angiography six months after the embolization.

Discussion

SDAVFs are predominantly found in the thoracolumbar region. The majority of SDAVF patients come to medical attention because of symptoms associated with myelopathy¹. SAH is extremely rare. There is only one case report on SDAVF of the lumbar spine presenting with SAH⁸. Although SDAVF in the cervical region is not found as frequently as in the cervical region, SAH as a presenting symptom is not as uncommon as was thought², in striking contrast to SDAVF located in the thoracolumbar re-

gion. To our knowledge, five cases of cervical SDAVF presenting with SAH have been reported³⁻⁷. It is unclear why this difference between locations exists in the frequency of SAH. As is the case with progressive myelopathy, SAH is believed to be caused by venous hypertension. Venous hypertension occurs when arterialized blood enters the medullary vein and reaches the valveless coronal venous plexus and radial vein⁹. The cephalad direction of the draining vein contributes to explain the frequency of SAH in cervical SDAVF.

Recently, NBCA embolization has been widely used as an initial treatment for SDAVF. With the aid of neurointerventional equipment advances, the technical feasibility of this approach is improving, and it offers the benefits of reduced invasiveness and earlier rehabilitation¹⁰. However, its relatively high recurrence rate might be a major weak point, though when NBCA can be clearly documented in the draining vein, the recurrence rate to zero. Of the five previously described cases of cervical SDAVF presenting with SAH, cure was achieved by surgical ligation in four (in one of our cases, initial attempts to embolize arterial feeders with NBCA failed). The remaining case, reported by Cahan et al³, was treated with a single detachable silicone balloon. This case was unusual in that the high-flow lesion was analogous to a single-hole vertebrovertebral fistula.

Figure 2 Post-treatment study. Anteroposterior (A) and lateral (B) angiograms of the left vertebral artery show SDAVF disappearance. Note NBCA: ethiodol mixture in the variceal draining vein and in the coronary venous system. C) T1-weighted MRI scan shows serpentine high signal intensities indicating the presence of NBCA: ethiodol mixture on the dorsal surface of the cervical cord.

Ours is the first case of a successful NBCA embolization of cervical SDAVF presenting with SAH. Our case had the common features of a cervical SDAVF, with the exception of the large varix of the draining vein. We tried to obtain NBCA penetration into both fistula and drain-

ing vein, which was eventually accomplished by multistep trials and with the help of fiber coils.

Conclusively, NBCA embolization is a good treatment choice for cervical SDAVF, even in cases presenting with SAH. Clinical and angiographic follow-up is recommended.

References

- 1 Berenstein A, Lasjaunias P: Surgical NeuroAngiography 5: Endovascular Treatment of Spine and Spinal Cord Lesions. Springer. Berlin 1992.
- 2 Kohno M, Takahashi H et Al: A cervical dural arteriovenous fistula in a patient presenting with radiculopathy: case report. *J Neurosurg* 84: 119-123, 1997.
- 3 Cahan LD, Higashida RT et Al: Variants of radiculomeningeal vascular malformations of the spine. *J Neurosurg* 66: 333-337, 1987.
- 4 Willinsky R, TerBrugge K et Al: The variable presentations of craniocervical and cervical dural arteriovenous malformations. *Surg Neurol* 34: 118-123, 1990.
- 5 Morimoto T, Yoshida S, Basugi N: Dural arteriovenous malformation in the cervical spine presenting with subarachnoid haemorrhage: case report. *Neurosurg* 31: 118-121, 1992.
- 6 Ikeda H, Fujimoto Y et Al: A rare case of high cervical spinal cord dural arteriovenous fistula presenting with intracranial subarachnoid haemorrhage. *No Shinkei Geka* 22: 1045-1048, 1994.
- 7 Do HM, Jensen ME, Cloft HJ, Kallmes DF, Dion JE: Dural Arteriovenous Fistula of the Cervical Spine Presenting with Subarachnoid Haemorrhage. *Am J Neuroradiol* 20: 348-350, 1999.
- 8 Koch C, Gottschalk S, Giese A: Dural arteriovenous fistula of the lumbar spine presenting with subarachnoid haemorrhage. Case report and review of the literature. *J Neurosurg* 100: 385-391, 2004.
- 9 McCutcheon IE, Doppman JL, Oldfield EH: Microvascular anatomy of dural arteriovenous abnormalities of the spine: a microangiographic study. *J Neurosurg* 84: 215-220, 1996.
- 10 Song JK, Gobin YP et Al: N-butyl 2-cyanoacrylate Embolization of Spinal Dural Arteriovenous Fistulae. *Am J Neuroradiol* 22: 40-47, 2001.

Taik-Kun Kim, M.D.
Department of Diagnostic Radiology
Korea University
College of Medicine, Ansan Hospital
#516 Kojan-Dong, Ansan City
Kyungki-Do 425-020, Korea
E-mail: tkkim@kumc.or.kr

EDITORIAL COMMENT

The authors present a very interesting case of arteriovenous lesion draining into the pial venous system of the upper cervical spinal cord and distally in the posterior fossa. As usual our capacity to differentiate between paraspinal AV shunt, dural shunt and radicular nerve AV shunt is difficult, particularly at the cervical level. In their case, the lesion does not opacify the epidural venous plexus, or the perivertebral venous plexus, therefore its location is likely to be either dural or radicular. Such situations have been encountered in some rare cases. Experience shows that these lesions are capable of producing either chronic myelopathy, as any slow flow spinal dural shunt or filum terminal shunt. In other situations at the upper cervical spine, and particularly at the foramen magnum, such radicular AVM or dural AV shunts can produce subarachnoid haemorrhage.

Such case must be stored in the memory as it certainly draws attention to the associated perineural venous enlargement during screening of subarachnoid haemorrhage. In such case careful opacification of the cervical artery should be performed.

Pierre Lasjaunias

Hôpital de Bicêtre
Service de Neuroradiologie Diagnostique
et Thérapeutique
Le Kremlin - Bicêtre, France