Case Report

Extensive Spinal Cord Infarction after Surgical Interruption of Thoracolumbar Dural Arteriovenous Fistula Presenting with Subarachnoid Hemorrhage

Sang-Hun Lee, M.D.,[1] Ki-Tack Kim, M.D.,[1] Sung-Min Kim, M.D.,[2] Dae-Jean Jo, M.D.[2]

Departments of Orthopaedic Surgery,[1] Neurosurgery,[2] Spine Center, Kyung Hee University East West Neomedical Center, Seoul, Korea

Nontraumatic intracranial subarachnoid hemorrhage (SAH) attributable to the thoracolumbar dural arteriovenous fistulas (DAVFs) has been extremely rare. A 41-year-old male patient was admitted with severe acute headache, neck stiffness, and pronounced low-back pain radiating to both legs. The T2-weighted MR imaging showed irregular signal void and enlarged, varix like pouch formation with spinal cord compression at the T11-12 level. The angiogram revealed a DAVF.
We report a DAVF case with SAH that revealed an extensive infarction from C5 to the conus medullaris after undergoing operative treatment.

KEY WORDS : Dural arteriovenous fistula · Subarachnoid hemorrhage · Spinal cord infarction.

INTRODUCTION

Dural arteriovenous fistula (DAVF, type I spinal arteriovenous malformation) is often developed at the thoracolumbar junction of middle-aged men with clinical symptoms including paraparesis, sensory deficits, and disturbance of bowel/bladder function resulted from venous hypertension and ischemic myelopathy[14,20,27]. There are also cases presenting with subarachnoid hemorrhage (SAH) when venous pressure is high[3,15]. Reports on nontraumatic intracranial SAH due to spinal DAVFs involving the cervical vertebra or the cervicothoracic junction have been made. To our knowledge, there is only one reported case of SAH occurred at the L4 level[15] and no study has described SAH located at the thoracolumbar junction.

Spinal cord infarction is a very rare disorder, accounting for less than 1% of all strokes. It's pathogenesis and prognosis remain largely unknown[18,19,31]. Some of the identified risk factors include reperfusion injury after aortic clamping

during thoracoabdominal aortic surgery[8,29,38], excessive hypoxia or systemic hypotension that sustained during operation[4,11,17,34], epidural anesthesia or lumbar puncture complications[6,9], unexpected embolic episode associated with embolization of the renal artery or mesenteric artery[5], cardiogenic thromboembolus, and artherosclerosis. In lower frequencies, cases with vascular diseases, polycythemia, decompression sickness, hypercoagulability, and meningeal infection have been reported[1,7,10,11,13,17].

In a meta-analysis of 44 spinal cord infarctions in 1996, Chesire et al.[7] reported 3 cases presenting with both spinal arteriovenous malformation and spinal cord infarction. However, the infarction extended from the cervical level to the thoracolumbar level was not found.

We report a DAVF case with SAH that revealed an extensive infarction from C5 to the conus medullaris after undergoing operation.

CASE REPORT

History. A 41-year-old male presented with severe acute headache, neck stiffness, and pronounced low-back pain radiating to both legs. He had intermittent both leg radiating pain during last two years. He had no history of diabetes, hypertension, smoking or coagulopathy.
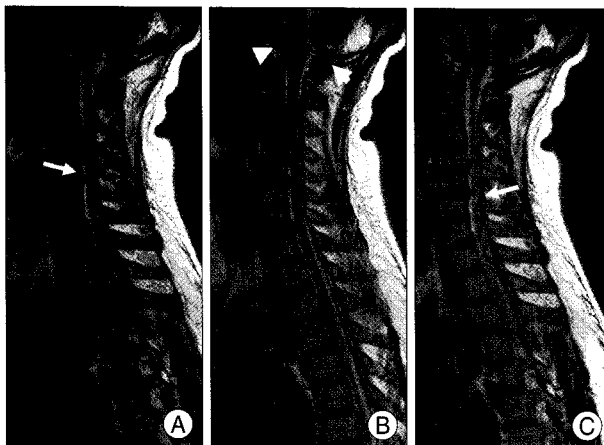
Examination. Neurological examination demonstrated

nuchal rigidity and a positive straight-leg raising sign. Laboratory study showed mild leukocytosis (14,500/μL), normal platelet count, prothrombin time (PT), and activated partial thromboplastin time (aPTT).

The T2-weighted whole spinal magnetic resonance imaging showed irregular signal void and enlarged, varix like pouch formation with spinal cord compression at the T11-12 level (Fig. 1). SAH was noted at the posterior cranial fossa and at the L5-S1 level. His cervical body to canal ratio at C5 was 0.45 and a narrow spinal canal from C3 to the thoracic spine was noted. High signal intensities were found at the C5-6 level with central disc herniation (Fig. 2). Under the impression of SAH associated with spinal DAVF, a spi-

nal angiogram was subsequently obtained. The angiogram revealed a DAVF with arterial blood supply from the segmental artery between T12 and L1 in an early phase and venous drainage into the intrathecal venous plexus involving varicose radiculospinal veins. But, the artery of Adamkiewicz (AKA) was not found (Fig. 3).

Operation. A midline posterior approach centered on the T12 region was performed with electrophysiological monitoring of somatosensory-evoked potential and motor-evoked potential. After laminectomy and vertical midline durotomy, exposed variably sized dilated tortous and serpentine perimedullary veins with the hematoma compressing the spinal cord. An enlarged fistula measured 2mm in diameter was located at the ventral side of the T12 root (Fig. 4). Temporary ligations were placed at the proximal end, interrupting the venous drainage of the fistula and causing the vein to
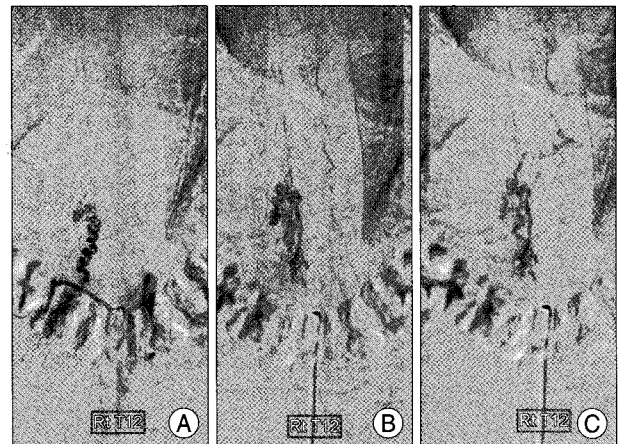

Fig. 1. Right paracentral (A) and midline (B) sagittal T2 images showing irregular fluid void and enlarged, varix like pouch formation (arrows) with spinal cord compression at the level T11-12. Collection of subarachnoid hemorrhage (arrow head) is noted at the level L5-S1.

Fig. 3. Anteroposterior angiogram revealing the dural arteriovenous fistula with arterial blood supply from the segmental artery between T12 and L1 in early phase (A) and venous drainage into the intrathecal venous plexus occurs via varicose radiculospinal veins (B and C).
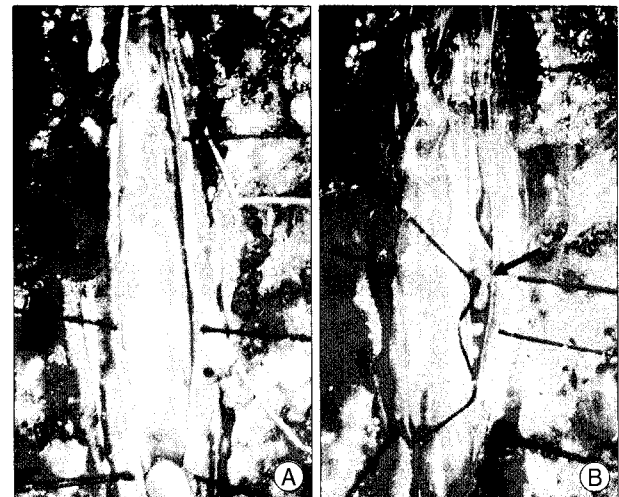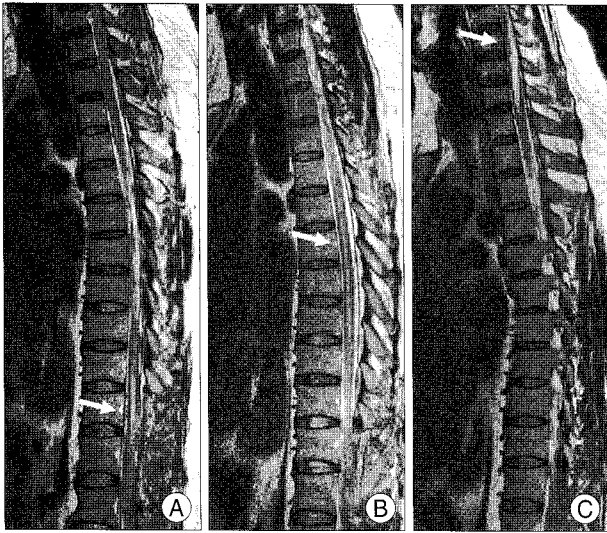

Fig. 2. Preoperative right paracentral (A), midline (B) and left paracentral (C) sagittal T2 images showing narrow spinal canal from C3 to thoracic spine. High signal intensities (arrows) are found on both paracentral images at the level C5-6 with central disc herniation. Subarachnoid hemorrhage within posterior cranial fossa is noted (arrow heads).

Fig. 4. Intraoperative photographs showing enlarged serpentine venous structures at the dorsal surface of spinal cord (A) and temporary ligation of fistula located just ventral side of T12 root.

Fig. 5-1. Immediate postoperative right paracentral (A), midline (B) and left paracentral (C) sagittal T2 images showing widespread hyperintensities (arrows) with cord swelling from the level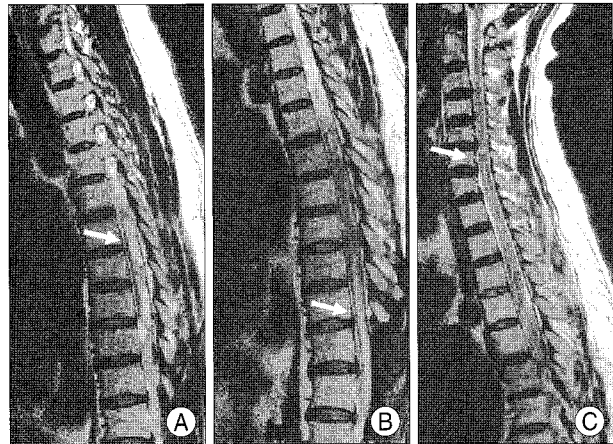 C5 to L1, consistent with spinal cord infarction. There is no residual arteriovenous fistula or varix like dilated vein.
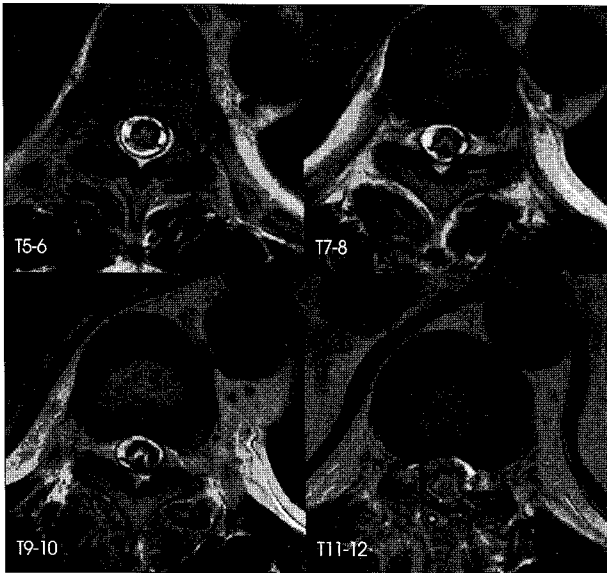
Fig. 5-2. Immediate postoperative axial T2 images showing bilateral hyperintensities corresponding to the whole gray matter.

Fig. 6. Eight months after surgery, the size and intensities of spinal cord infarction (arrows) are decreased but still visible on right paracentral (A), midline (B) and left paracentral (C) sagittal T2 images.

turn blue. There was no specific change on the electrophysiological monitoring for 20 minutes. Then, the lesion was coagulated and ligated. The dura mater and surgical wounds were closed. Time for operation was 200 minutes and intraoperative blood loss was 450 mL. The lowest blood pressure recorded was 90/60 mmHg, lasting about 20 minutes, which promptly recovered after volume expansion.

The immediate postoperative course was uneventful and the patient had grossly normal motor functions. The patient's neurological examination revealed paraparesis (grade III-IV) after 4 hours following the ligation. Under the impression of epidural hematoma, we decided to perform emergent decompressive surgery. However, there was no evidence of hematoma in the epidural space, we opened the durotomy site for exploration. Severe cord swelling was seen. Thus, we carried out extended decompressive laminectomy with duroplasty using a dural substitute (Neuropatch : B. Braun Melsungen AG, Melsungen, Germany), and placed a lumbar CSF drain. The patient exhibited a complete paraplegia after the second operation.

Postoperative Course. Spinal MR imaging was obtained postoperatively, which confirmed spinal cord infarction from C5-6 to the conus medullaris (Fig. 5-1, 2). During the first 3 months after operation, neurological deficits improved gradually and reached a plateau in recovery. At the 1-year clinical follow-up examination, the patient still complained of right leg weakness (grade IV) and disturbance of bowel and bladder function. He could ambulate with support of a cane. Repeated MR imaging 8 months later showed decreased size and intensities of spinal cord infarction, but hyperintensities were still visible (Fig. 6).

DISCUSSION

The anterior spinal artery is the one involved with the pathogenic mechanism in spinal cord infarction[35]. The most important feeder among them is the AKA, which branches between T9 and T12 on the left side in 65-75%. Interruption of blood supply due to embolism or injury of the artery has been cited as the main cause of spinal cord infarction[24,26].

Spinal cord infarction is also referred to as acute spinal cord ischemia syndrome (ASCIS)[22,30]. The general factors of ischemia include global hypoperfusion, reperfusion injury, coagulopathy, vasculitis, and decompression sickness

while the local factors are epidural hematoma and thromboembolic accident.

Our case showed extensive infarction reaching the cervical cord without notable risk factors after surgical interruption of thoracolumbar DAVF. However, this patient had some features to note : 1) the cervical neural canal was so narrow that preoperative MR imaging showed a nearly obliterated CSF space surrounding the spinal cord, 2) a high signal intensity resulted from the central disc herniation at C5-6 was observed on T2W sagittal MR imaging, 3) SAH extended to the posterior cranial fossa and the sacral area, and 4) a numbder of enlarged, dilated and tortuous epidural veins were observed during operation. These four features led us to presume that the patient had high intradural pressure within a relatively narrow neural canal.

Considering the above features, we reached the following two pathogenic mechanisms for the extensive infarction :

1) The blood vessel from which DAVF originated might have been the AKA. Preoperative angiography confirmed the feeding site on the right, but the AKA was not found on the left. Although the DAVF originated from the intervertebral foramen between T12 and L1 on the right side, the AKA may extend to anywhere between T5 and L2. About 25% of the AKA is known to extend on the right side[16,23]. Hemodynamic changes or thrombosis in the spinal cord caused by the interruption of DAVF originating from the AKA might have resulted in the extensive infarction. However, this possibility alone does not clearly explain the infarction extending from the upper thoracic to the cervical cord.

2) The next possible mechanism is the rapid increase of blood flow in the spinal cord that had been under the ischemic state in an abnormal CSF dynamic pathway within a narrow and high-pressured spinal canal when DAVF was interrupted. As a result, reperfusion injury might have been sustained, leading to secondary injury and swelling of the spinal cord.

Chronic spinal cord compression is known to cause subarachnoid fibrosis, reducing collateral blood flow and the compensation mechanism for ischemic events[36]. In fact, there are reports on the cases that developed spinal cord infarction due to compression caused by cervical myelopathy[2,25,32]. In our case, the narrow spinal canal is considered to have played a major role in developing extensive infarction.

According to recent studies, a severely compressed cervical spinal cord due to cervical spondylotic myelopathy accompanies significant swelling after decompression[21,37]. There are also studies that suggest the transient root palsy observ-

ed after decompressive surgery is an incomplete cord injury caused by a reperfusion injury[29,37]. This increases the possibility of reperfusion injury and secondary infarction resulting from the temporarily increasing arterial pressure when the fistula shunt was blocked.

The prognosis for spinal cord infarction remains unclear, but the early neurologic symptoms and age of a patient have been cited as major factors that affect prognosis[7,11,20,23]. Our patient showed gradual recovery of motor and sensory functions after operation, but partial paralysis persisted on the right lower limb. In addition, bladder and bowel functions have not recovered, showing a severe sequale.

CONCLUSION

Based on this case report, we recommend that the risk of neurologic complications be considered in advance when performing operation on a DAVF patient with a narrow spinal canal. Surgeons are also advised to check the status of blood supply to the spinal cord by confirming the AKA of the spinal cord and the feeding artery on preoperative angiography, in an effort to predict and prevent postoperative complications.

References

1. Almasanu BP, Owensby JR, Pavlakis SG, Edwards JH : Spinal cord infarction in meningitis : polygenic risk factors **Pediatr Neurol 32** : 124-126, 2005
2. Arai S, Utsunomiya H, Wakugawa Y, Uwadoko T : [A case of spinal cord infarction caused by cervical disc herniation.] **Brain Nerve 59** : 997-1000, 2007
3. Aviv RI, Shad A, Tomlinson G, Niemann D, Teddy PJ, Molyneux AJ, et al. : Cervical dural arteriovenous fistulae manifesting as subarachnoid hemorrhage : report of two cases and literature review. **AJNR Am J Neuroradiol 25** : 854-858, 2004
4. Bromage PR : Letter : Spinal thrombophlebitis after hypotensive anaesthesia. **N Z Med J 80** : 519-520, 1974
5. Burbank SA, Vaccaro AR, Goins ML, Tribus CB, Manon-Espaillat R : Thoracic paraparesis following an embolic vascular event during lumbar spinal surgery. **J Spinal Disord Tech 19** : 68-72, 2006
6. Chan LL, Kumar AJ, Leeds NE, Forman AD : Post-epidural analgesia spinal cord infarction : MRI correlation. **Acta Neurol Scand 105** : 344-348, 2002
7. Cheshire WP, Santos CC, Massey EW, Howard JF Jr : Spinal cord infarction : etiology and outcome. **Neurology 47** : 321-330, 1996
8. Coselli JS, LeMaire SA, Miller CC 3rd, Schmittling ZC, Köksoy C, Pagan J, et al. : Mortality and paraplegia after thoracoabdominal aortic aneurysm repair : a risk factor analysis. **Ann Thorac Surg 69** : 409-414, 2000
9. Dahlgren N, Törnebrandt K : Neurological complications after anaesthesia. A follow-up of 18,000 spinal and epidural anaesthetics performed over three years. **Acta Anaesthesiol Scand 39** : 872-880, 1995
10. Dawson DM, Potts F : Acute nontraumatic myelopathies. **Neurol Clin 9** : 585-603, 1991
11. Duggal N, Lach B : Selective vulnerability of the lumbosacral spinal cord after cardiac arrest and hypotension. **Stroke 33** : 116-121, 2002
12. Feasby TE, Ferguson GG, Kaufman JC : Isolated spinal cord arteritis.

Can J Neurol Sci 2 : 143-146, 1975
13. Galetta SL, Balcer LJ, Lieberman AP, Syed NA, Lee JM, Oberholtzer JC : Refractory giant cell arteritis with spinal cord infarction. Neurology 49 : 1720-1723. 1997
14. Kinouchi H, Mizoi K, Takahashi A, Nagamine Y, Koshu K, Yoshimoto T : Dural arteriovenous shunts at the craniocervical junction. J Neurosurg 89 : 755-61, 1998
15. Koch C, Gottschalk S, Giese A : Dural arteriovenous fistula of the lumbar spine presenting with subarachnoid hemorrhage. Case report and review of the literature. J Neurosurg 100 : 385-391, 2004
16. Koshino T, Murakami G, Morishita K, Mawatari T, Abe T : Does the Adamkiewicz artery originate from the larger segmental artery? J Thorac Cardiovasc Surg 117 : 898-905, 1999
17. Lynch DR, Dawson TM, Raps EC, Galetta SL : Risk factors for the neurologic complications associated with aortic aneurysms. Arch Neurol 49 : 284-288, 1992
18. Martinelli V, Comi G, Rovaris M, Filippi M, Colombo B, Locatelli T, et al. : Acute myelopathy of unknown aetiology : a clinical, neurophysiological and MRI study of short- and long-term prognostic factors. J Neurol 242 : 497-503, 1995
19. Masson C, Pruvo JP, Meder JF, Cordonnier C, Touzé E, De La Sayette V, et al. : Spinal cord infarction : clinical and magnetic resonance imaging findings and short term outcome. J Neurol Neurosurg Psychiatry 75 : 1431-1435, 2004
20. Merland JJ, Riche MC, Chiras J : Intraspinal extramedullary arteriovenous fistulae draining into the medullary veins. J Neuroradiol 7 : 271-320, 1980
21. Nagashima H, Morio Y, Teshima R : Re-aggravation of myelopathy due to intramedullary lesion with spinal cord enlargement after posterior decompression for cervical spondylotic myelopathy : serial magnetic resonance evaluation. Spinal Cord 40 : 137-141, 2002
22. Nedeltchev K, Loher TJ, Stepper F, Arnold M, Schroth G, Mattle HP, et al. : Long-term outcome of acute spinal cord ischemia syndrome. Stroke 35 : 560-565, 2004
23. Nojiri J, Matsumoto K, Kato A, Miho T, Furukawa K, Ohtsubo S, et al. : The Adamkiewicz artery : demonstration by intra-arterial computed tomographic angiography. Eur J Cardiothorac Surg 31 : 249-255, 2007
24. Novy J, Carruzzo A, Maeder P, Bogousslavsky J : Spinal cord ischemia : clinical and imaging patterns, pathogenesis, and outcomes in 27 patients. Arch Neurol 63 : 1113-1120, 2006
25. Odaka M, Hirata K : [Anterior spinal artery syndrome due to cervical spondylosis presenting as cervical angina.] No To Shinkei 56 : 961-963, 2004
26. Romanes GJ : The arterial blood supply of the human spinal cord.

Paraplegia 59 : 199-207, 1965
27. 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
28. Rosenthal D : Spinal cord ischemia after abdominal aortic operation : is it preventable? J Vasc Surg 30 : 391-397, 1999
29. Sakaura H, Hosono N, Mukai Y, Ishii T, Yoshikawa H : C5 palsy after decompression surgery for cervical myelopathy : review of the literature Spine (Phila Pa 1976) 28 : 2447-2451, 2003
30. Salvador de la Barrera S, Barca-Buyo A, Montoto-Marqués A, Ferreiro-Velasco ME, Cidoncha-Dans M, Rodriguez-Sotillo A : Spinal cord infarction : prognosis and recovery in a series of 36 patients. Spinal Cord 39 : 520-525, 2001
31. Sandson TA, Friedman JH : Spinal cord infarction. Report of 8 cases and review of the literature. Medicine (Baltimore) 68 : 282-292, 1989
32. Sayer FT, Vitali AM, Low HL, Paquette S, Honey CR : Brown-Sèquard syndrome produced by C3-C4 cervical disc herniation : a case report and review of the literature. Spine (Phila Pa 1976) 33 : E279- E282, 2008
33. Seichi A, Takeshita K, Kawaguchi H, Nakajima S, Akune T, Nakamura K : Postoperative expansion of intramedullary high-intensity areas on T2-weighted magnetic resonance imaging after cervical laminoplasty. Spine (Phila Pa 1976) 29 : 1478-1482; discussion 1482, 2004
34. Singh U, Silver JR, Welply NC : Hypotensive infarction of the spinal cord. Paraplegia 32 : 314-322, 1994
35. Sliwa JA, Maclean IC : Ischemic myelopathy : a review of spinal vasculature and related clinical syndromes. Arch Phys Med Rehabil 73 : 365-372, 1992
36. Yoshizawa H : Presidential address : pathomechanism of myelopathy and radiculopathy from the viewpoint of blood flow and cerebrospinal fluid flow including a short historical review. Spine (Phila Pa 1976) 27 : 1255-1263, 2002
37. Yukawa Y, Kato F, Ito K, Horie Y, Hida T, Machino M, et al. : Postoperative changes in spinal cord signal intensity in patients with cervical compression myelopathy : comparison between preoperative and postoperative magnetic resonance images. J Neurosurg Spine 8 : 524-528, 2008
38. Wan IY, Angelini GD, Bryan AJ, Ryder I, Underwood MJ : Prevention of spinal cord ischaemia during descending thoracic and thoracoabdominal aortic surgery. Eur J Cardiothoracic Surg 19 : 203-213, 2001