CASE REPORT

J Korean Neurosurg Soc 38 : 74-76, 2005

A Case of Spinal Intradural Extramedullary Cavernous Angioma

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Cavernous angiomas represent 5 to 12% of spinal vascular malformations and usually are located at the vertebral body level with possible extension into the extradural space. The intradural intramedullary cavernous angioma occurs in about 3% of cases, whereas extramedullary localization is extremely rare. We report a case of intradural extramedullary cavernous angioma in which the patient presented with low back pain and both leg pain. The magnetic resonance imaging study showed intraspinal mass lesion at L1-2. It was removed totally through laminectomy of L1-2 and confirmed as cavernous angioma. The postoperative course was uneventful without any neurologic deficit. We report this unusual spinal malformation.

KEY WORDS : Cavernous angioma · Intradural-extramedullary.

Introduction

Cavernous angiomas are uncommon vascular malformations composed of vascular spaces lined with a single endothelial cell layer with no interposed neural tissues. Cavernous angiomas can occur anywhere in the neuraxis and account for 5~12% of all vascular lesions of the spine[3,5,8,12]. In the spinal region, cavernous angiomas are most common in the vertebral body with occasional extension into the extradural space. Approximately 3% of spinal cavernous angiomas are intradural, usually with an intramedullary location[2,4,6,9,11,12]. Intradural extramedullary cavernous angiomas are not common. We present a case of an intradural extramedullary cavernous angioma of the lumbar region.

Case Report

A 52-year-old man was admitted with severe low back and both leg pain for 2 years. These symptoms were aggravated 1month prior to admission. A neurological examination revealed mild paresthesia on the right leg and no motor weakness. There were no pathologic reflexes. Laboratory findings were normal. Plain radiographs of his spine were normal.

Magnetic resonance(MR) imaging of the spine showed a heterogenous non-enhancing mass obliterating the spinal canal at the L2 level. The lesion exhibited a mixed signal intensity on T1- and T2- weighted images and was surrounded by dark hemosiderin ring suggesting multiple stage hemorrhage (Fig. 1). At operation, a L1-L2 laminectomy was performed. The dura was not tense. When the dura was opened under the microscope, a dark red lesion with clotted blood was readily identified at the end of the conus medullaris which extended upward (Fig. 2). It adhered to the nerve roots. The nerve roots adherent to the mass were carefully dissected. The lesion seemed to arise primarily from the nerve root. Gross total resection was ultimately achieved with sacrifice of 1 nerve root.

The histological examination revealed an irregular, dilated, sinusoidal space lined with a single layer of endothelial cells (Fig. 3). This was surrounded by hematoma. These findings were consistent with those of a cavernous angioma. After the operation, the patient's pain quickly resolved.

Discussion

Cavernous angiomas or cavernomas are uncommon vascular lesions of the CNS, considered to be malformations, and are clearly differentiated from vascular neoplasms[1-4]. Cavernous angiomas may occur singly or in clusters within

• Received : December 9, 2004 • Accepted : February 21, 2005

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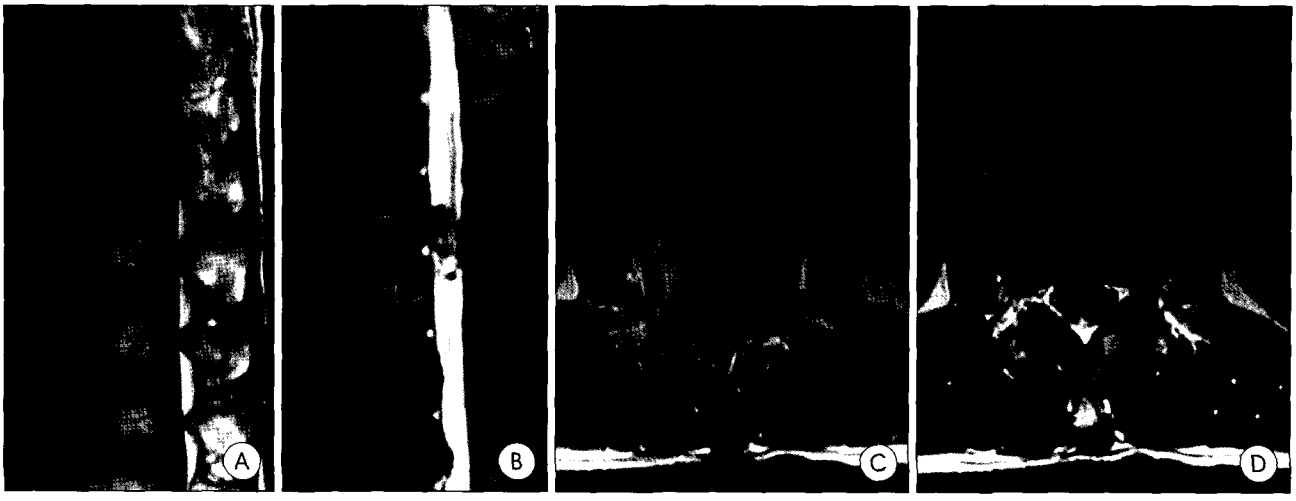

Fig. 1. Preoperative magnetic resonance images showing a heterogeneous non-enhancing mass obliterating the spinal canal at the L2 level. The lesion exhibited a mixed signal intensity on T1(A, C) and T2(B, D) weighted images and was surrounded by dark hemosiderin ring suggesting multiple stage hemorrhage.

Fig. 2. Intraoperative photograph demonstrating a dark red mass(white arrow head) adherent to the nerve root (white arrow).

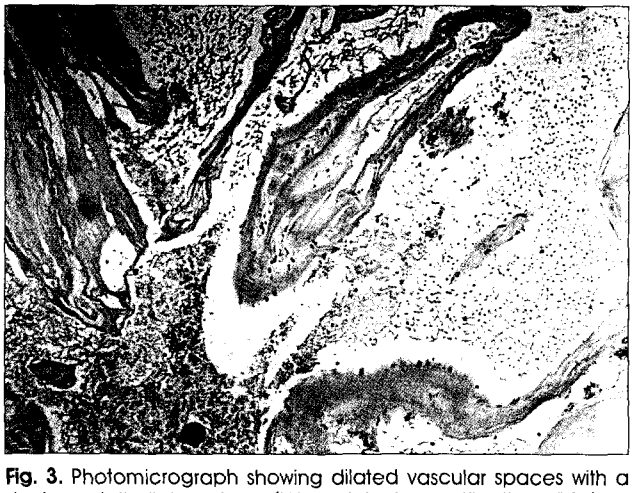

Fig. 3. Photomicrograph showing dilated vascular spaces with a single endothelial cell layer (H&E, original magnification×100).

the central nervous system. They are more common in the brain than in the spine. They constitute 5 to 18% of intracranial vascular malformations and 5 to 12% of the spinal ones. The great majority of spinal cavernomas are located in the vertebral bodies with occasional extension into the extradural space but pure epidural lesions are uncommon[3,5,7,8,12]. Approximately 3% of spinal cavernous angiomas are intradural, usually with an intramedullary. Intradural extramedullary lesions are extremly rare of all cavernous malformations[2,4,6,9,11,12].

Table 1 provides a summary if our case and the other 22 surgically treated cases. In the spinal intradural extramedullary space, hemangiomas may arise from the blood vessels of the nerve roots, inner surface of the dura, and the pial surface of the spinal cord. The frequent intraoperative finding that the lesion adheres to the nerve roots or the spinal cord suggests that it arises from the vessels on the surface of the roots or spinal cord and extends into the intradural sub- or epiarachnoid space. The relatively high associated incidence of SAH compared with other intradural extramedullary lesions may be explained by the vascular nature and restricted mobility by the nerve roots in the dynamic vertebral canal[8].

The MRI is the diagnostic modality of choice, and spinal angiography shows negative results. The MR imaging findings depends on the stage of hemorrhage within the lesion. The typical MRI appearance is that of a well-defined lesion with hypointense areas intermixed with small punctate areas of hyperintensity on both T1- and T2-weighted images. They are generally surrounded by a hypointense hemosiderin ring on T2-weighted imaging. They are enhanced variably. There is typically no signal void which is characteristically seen in high flow vascular lesions[4].

The optimal therapy for spinal intradural extramedullary cavernomas is surgery. Macroscopically the lesion is well demarcated and total removal is possible in most cases and allows preservation of normal neural tissues, although the lesion is usually attached to the nerve roots or the spinal cord.

Because the lesion tends to bleed and the patient's clinical

Table 1. Summary of cases involving surgically treated intradural extramedullary cavenous angiomas*

Authors & year	Age (yrs.) sex	Location	Extent of resection	Lesion origin	Outcome
Poger, et al., 1951	22, F	T-11	total excision	ND	worse
Floris, 1958	57, M	T-12	total excision	ND	ND
Hirsch, et al., 1965	20, M	L2-3	total	root	incomplete recovery
Pansini & Lo Re, 1966	46, M	L-2	total	root	incomplete recovery
Ortner, et al., 1973	22, M	C4-7	total	root	no improvement
Heimberger, et., 1982	24, M	T2-3	total	root	excellent
Ueda, et al., 1987	28, M	L1-2	total	root	excellent
Pagni, et al., 1990	26, M	T12-L1	total	root	excellent
Ramos, et al., 1990	67, F	L-3	total	filum terminale	excellent
Mastronardi, et al., 1992	49, F	T-4	total	root	excellent
Mori, et al., 1991	65, M	T-1	total	spinal cord	excellent
Acciarri, et al., 1992	54, F	C2-3	total	dura mater	excellent
Shgarma, et al., 1992	63, M	T-12	total	root/spinal cord	no improvement
	43, M	T-5	total	root/spinal cord	excellent
Bruni, et al., 1994	28, M	L-2	total	root	excellent
Cervoni, et al., 1995	26, F	L1-2	total	root	excellent
	32, M	L-5	total	root	incomplete recovery
Rao. et al.. 1997	60, M	L1-3	total	root	excellent
	35, F	T-12	subtotal	spinal cord	no improvement
Duke, et al., 1998	49, F	L-4	total	root	excellent
Kim, et al 2001	65, M	L4	total	root	excellent
Nozaki, et al 2003	51, M	C5-6	total	root/dentate ligament	excellent
present case	52, M	L2	total	root	excellent

* ND = not described

outcome depends on the severity of the preoperative neurological status, surgical treatment should be performed before the patient's neurological deficits become irreversible[8].

The microsurgical technique makes the dissection and the removal of the vascular malformation less difficult.

Conclusion

An intradural extramedullary cavernous angioma is a rare vascular malformation, consisting of vascular spaces similar to those of sinusoids and lined with a single layer of endothelial cells without any interposed glial or nervous tissue. There are only 20 surgically treated cases reported in the literature[8,10]. We report the case of a patient with a lumbar intradural extramedullary cavernous angioma on whom surgery was performed.

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