J Korean Neurosurg Soc 48: 272-275, 2010

Case Report

# Capillary Hemangioma of the Thoracic Spinal Cord

Sung-Kyun Chung, M.D., Taek-Kyun Nam, M.D., Seung-Won Park, M.D., Sung-Nam Hwang, M.D.

Department of Neurosurgery, Chung-Ang University Yongsan Hospital, Seoul, Korea

Capillary hemangiomas are common soft tissue tumors on the skin or mucosa of the head and neck in the early childhood, but very rare in the neuraxis. A 47-year-old man presented with one month history of back pain on the lower thoracic area, radiating pain to both legs, and hypesthesia below T7 dermatome. Thoracic spine MRI showed 1×1.3×1.5 cm, well-defined intradural mass at T6-7 disc space level, which showed isointensity to spinal cord on T1, heterogeneous isointensity on T2-weighted images, and homogeneous strong enhancement. The patient underwent T6-7 total laminotomy, complete tumor removal and laminoplasty. Histologically, the mass showed a capsulated nodular lesion composed of capillary-sized vascular channels, which were tightly packed into nodules separated by fibrous septa. These features were consistent with capillary hemangioma.

**KEY WORDS**: Capillary hemangioma · Spinal cord · Intradural extramedullary tumor.

## INTRODUCTION

Capillary hemangioma (CH) is a benign tumor or tumorlike lesion most often encountered on the skin and in the soft tissues<sup>3)</sup>. It consists of aggregation of abnormal blood vessels that used to be better known by the misnomer 'pyogenic granuloma<sup>24</sup>. Within central nervous system (CNS), especially in the spinal cord, this tumor very rarely occurs. Spinal intradural CH shows isointensity relative to spinal cord on T1, isointensity or slight hyperintensity on T2-weighted images, and homogeneous enhancement on MRI<sup>17)</sup>. Because these MRI findings are similar or sometimes same as those of other common intradural extramedullary spinal tumors such as meningioma, neurofibroma and schwannoma, it is often difficult to differentiate intradural CH from other IDEM (Intradural extramedullary) tumors by MRI<sup>12,14)</sup>.

We report a rare case of a spinal intradural extramedullary capillary hemangioma on the thoracic spinal cord.

# **CASE REPORT**

A 47-year-old man presented with 1-month history of

Received: August 3, 2009 • Revised: May 27, 2010

Accepted: August 9, 2010

Tel: +82-2-748-9872, Fax: +82-2-798-2818

E-mail: tarheelk@hanmail.net

back pain on the lower thoracic area and radiating pain down to both legs. On neurologic examination, he demonstrated sensory impairment below T7 dermatome, increased deep tendon reflex and ankle clonus bilaterally. Thoracic spine MRI showed  $1 \times 1.3 \times 1.5$  cm sized, well-defined intradural mass at T6-7 disc space level, which compressed and displaced spinal cord ventrolaterally. This lesion was isointense to spinal cord on T1-weighted images and heterogeneously isointense on T2-weighted images with homogeneous strong enhancement (Fig. 1). The patient underwent T6-7 total laminectomy. On opening the dura, a round, wellcircumscribed and dark reddish mass was seen on the spinal cord, which was easily dissected from the dura. Under microscope, the mass was dissected out from the spinal cord successfully though part of it was strongly adhered to the spinal cord. After complete removal, we carried dural closure and T6-7 laminoplasty. The patient's symptoms disappeared after the surgery. Sensory impairment was gradually improved on follow-up after discharge.

On histological examination, the mass had a lobular, capillary, and hypercellular structure which was separated by fibrous connective tissue band and was composed of numerous, tightly packed, capillary size vessels that were lined by a single layer of cytologically benign endothelial cells. Blood vessels varied widely in size from small lumina lined with endothelial cells to dilated vessels lined with flattened endothelium. Many scattered stromal lymphocytes were present. All of these features were consistent with a capillary hemangioma (Fig. 2).

Address for reprints : Sung-Nam Hwang, M.D., Ph.D. Department of Neurosurgery, Chung-Ang University Yongsan Hospital, Hangang-ro 3-ga, Yongsan-gu, Seoul 140-757, Korea

#### DISCUSSION

Spinal cord tumors account for about 15% of all central nervous system (CNS) neoplasm. Vascular lesions comprise about 6-7% of all spinal intradural tumors<sup>13)</sup>. Spinal vascular tumors may be classified as capillary telangiectasias, cavernous angioma, capillary hemangiomas, arteriovenous malformations or venous malformations<sup>16)</sup>. The neuro-epithelium, ontogenetically giving rise to the distal spinal cord, is of mesodermal origin. Frequently, tumors at the level of the conus medullaris and cauda equina contain mesodermal elements. The occurrence of vascular lesions involving the conus medullaris and cauda roots in a metameric distribution has occasionally been recognized.

Capillary hemangioma, one of the spinal vascular tumors, is characterized by a lobular architecture, with each lobule separated by septa of fibrous connective tissue and consisting of a myriad of small and very small capillaries lined by endothelial cells<sup>4</sup>). Because it is usually well demarcated from the surrounding parenchyma by a connective tissue capsule and reveals mild to moderate mitotic activity, CH can be classified into a benign vascular tumor or tumor-like lesion, despite the lack of precise understanding of the details of its development and growth<sup>4,14,15</sup>).

Spinal intradural CH are commonly presented in their fifth to sixth decades of life<sup>13)</sup>. There is no obvious sex predilection. It usually presents at the sur-

face of the distal spinal cord<sup>15,17)</sup>, close to the conus medullaris or attached to nerve roots of the cauda equina<sup>5,7,12,14,17)</sup>. Within the neuraxis, only a few cases of CH in the meninges, cauda equina, epidural, or intramedullary locations have been reported (Table 1).

Although clinical symptoms of CH are similar to these of other intramedullary tumors, CH infrequently shows severe neurological deficits at diagnosis due to the discrete and noninfiltrative nature.

Spinal MR findings of spinal intradural extramedullary

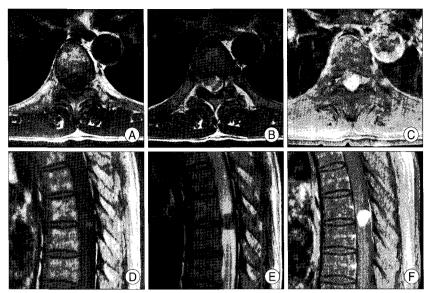
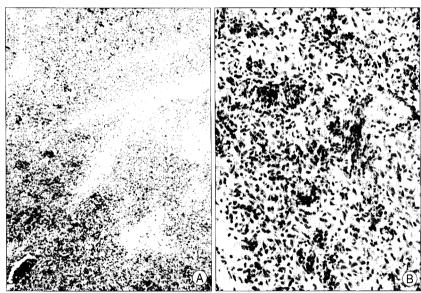


Fig. 1. Pre-operative spine magnetic resonance (MR) images. Thoracic spine MR scans show 1 × 1.3 × 1.5 cm sized, well-defined and intradural mass at T6-7 level, which compresses and displaces spinal cord ventrolaterally. The mass shows isointensity on T1-weighted images (A and D), heterogenous intensity on T2-weighted images (B and E) and homogeneous strong enhancement (C and F).



**Fig. 2.** Photographs of the capillary hemangioma. A: Lobular, capillary and hypercellular structure separated by fibrous tissue septa and fed by arteries (H&E, original magnification × 100). B: Blood vessels vary in size from small lumina lined with endothelial cells to dilated vessels lined with flattened endothelium. Many scattered stromal lymphocytes are present (H&E, original magnification × 400).

CH are characterized by isointensity or slightly hyperintensity relative to spinal cord on T1, isointensity or slightly hyperintensity on T2-weighted images, and homogeneous enhancement<sup>5,7,12,14,15)</sup> as in our patient. The differential diagnosis for such enhancing intradural extramedullary lesions in the spinal canal includes meningioma, hemangioma, schwannoma, hemangioblastoma, and paraganglioma. These signal characteristics may help in differentiating CH from meningioma, a much more common intradural extramedullary tumor, as meningiomas display low- or isointensity on T2-weighted

Table 1. Summary of previously reported cases of spinal intradural capillary hemangiomas

No	Author, year	Case	Sex (n)/Age	Location
1	Hanakita et al. <sup>5)</sup>	1 case	M/58	L1-L2
2	Mastronardi et al. <sup>11)</sup>	1 case	M/41	L5
3	Zander et al. <sup>19)</sup>	1 case	F/51	L4-L5
4	Holtzman et al.7)	1 case	F/56	1.4
5	Roncaroli et al.14)	9 cases	M (6) & F (3)/40-62	T5 (1), Cauda equina (8)
6	Nowak et al. <sup>12)</sup>	1 case	F/63	T12-L1
7	Shin et al. <sup>22)</sup>	1 case	F/66	T8-T9
8	Roncaroli et al. <sup>15)</sup>	4 cases	M (3) & F (1)/42-64	T10-T11 (3), Conus (1)
9	Abdullah et al.1)	1 case	F/32	T9-T10
10	Abe et al.2)	7 cases	M (7)/43-80	T7-T11 (7)
11	Kim et al. <sup>10)</sup>	1 case	M/59	L1-L2
12	Our study	1 case	M/47	T6-7

M : male, F : female

images. However, these are similar to those of intradural extramedullary neurogenic tumors, such as schwannoma, so differentiating between the two is difficult. Cavernous angiomas also have similar MR findings with CH. These account for 5-12% of all vascular space-occupying masses of the spine and are most frequently at the level of the conus medullaris and cauda equina, which clearly favor the cerebral hemispheres, but may occur anywhere in the neuraxis<sup>8,13)</sup>. Because cavernous angioma has a recurrent bleeding tendency, it often presents with acute symptoms and frequently reveals thrombosis and perivascular hemosiderin deposit, which result in heterogeneous high and low signal intensity consistent with subacute and chronic hemorrhage on MR<sup>18)</sup>. The identification of draining veins on MR images may help in differentiating the hypervascular lesions such as hemangioma, hemangioblastoma, and paraganglioma from the other nonhypervascular lesions and suggest the need for preoperative angiography and embolization.

Histologically, other spinal tumors except spinal vascular lesions can be definitely differentiated. Among the spinal vascular tumors, cavernous angioma and CH can be differentiated with other vascular tumors such as arteriovenous malformation and capillary telangiectasia which lack the distinct demarcation from the surrounding spinal cord parenchyma and invariably feature a racemose histological architecture, with isles of parenchyma between the vascular channels<sup>9</sup>. Cavernous angioma and CH have similar histological features of hemangiomas. Hemangioma can be divided into cavernous and capillary type depending on the dominant vessel size at microscopy. Cavernous angiomas are comprised of closely packed, large dilated vascular channels without interposed neural tissue<sup>8,16)</sup>. Unlike cavernous angioma, capillary angiomas are characterized by a myriad of small and very small capillaries lined by endothelial cells8. However, CH can show a more or less cavernous component, which means transitional forms<sup>6,14)</sup>.

A reasonable therapeutic strategy of CH is, as with other architecturally solid lesions, complete surgical resection and follow-up imaging study.

### **CONCLUSION**

We experienced a rare intradural extramedullary vascular tumor of the thoracic spine and performed surgical excision. After the operation, the patient's symptoms were improved. Histological diagnosis was capillary hemangioma. We suggest it should be considered in the differential diagnosis of intradural extramedullary spinal cord tumors.

#### References

- Abdullah DC, Raghuram K, Phillips CD, Jane JA Jr, Miller B: Thoracic intradural extramedullary capillary hemangioma. AJNR Am J Neuroradiol 25: 1294-1296, 2004
- Abe M, Tabuchi K, Tanaka S, Hodozuka A, Kunishio K, Kubo N, et al.: Capillary hemangioma of the central nervous system. J Neurosurg 101: 73-81, 2004
- 3. Abe M, Misago N, Tanaka S, Masuoka J, Tabuchi K: Capillary hemangioma of the central nervous system: a comparative study with lobular capillary hemangioma of the skin. Acta Neuropathol 109: 151-158, 2005
- Enzinger FM, Weiss SW: Benign tumors and tumor-like lesions of blood vessels in soft tissues tumors: soft tissue tumors, ed 3. St. Louis: Mosby, 1995, pp579-626
- Hanakita J, Suwa H, Nagayasu S, Suzuki H: Capillary hemangioma in the cauda equina: neuroradiological findings. Neuroradiology 33: 458-461, 1991
- Hida K, Tada M, Iwasaki Y, Abe H: Intramedullary disseminated capillary hemangioma with localized spinal cord swelling: case report. Neurosurgery 33: 1099-1101, 1993
- Holtzman RN, Brisson PM, Pearl RE, Gruber ML: Lobular capillary hemangioma of the cauda equina. Case report. J Neurosurg 90: 239-241, 1999
- Jellinger K: Pathology of spinal vascular malformations and vascular tumors: spinal angiomas advances in diagnosis and therapy. Berlin: Springer-Verlag, 1978, pp184
- Kalimo H, Kaste M, Haltia M: Vascular diseases in Graham DI, Lantos PI (eds): Greenfield neuropathology, ed 6. London: Arnold Publishing, 1997, pp315-396

- Kim KJ, Lee JY, Lee SH: Spinal intradural capillary hemangioma. Surg Neurol 66: 212-214, 2006
- Mastronardi L, Guiducci A, Frondizi D, Carletti S, Spera C, Maira G: Intraneural capillary hemangioma of the cauda equina. Eur Spine J 6: 278-280, 1997
- 12. Nowak DA, Gumprecht H, Stölzle A, Lumenta CB: Intraneural growth of a capillary haemangioma of the cauda equina. Acta Neurochir (Wien) 142: 463-467; discussion 467-468, 2000
- 13. Nowak DA, Widenka DC: Spinal intradural capillary haemangioma: a review. Eur Spine J 10: 464-472, 2001
- 14. Roncaroli F, Scheithauer BW, Krauss WE: Hemangioma of spinal nerve root. J Neurosurg (Spine 2) 91: 175-180, 1999
- 15. Roncaroli F, Scheithauer BW, Krauss WE: Capillary hemangioma of

- the spinal cord. Report of four cases. J Neurosurg 93: 148-151, 2000
- Russel DS, Rubinstein LJ: Pathology of tumors of the nervous system, ed 5. Baltimore: William & Wilkins, 1989, pp730-736
- Shin JH, Lee HK, Jeon SR, Park SH: Spinal intradural capillary hemangioma: MR findings. AJNR Am J Neuroradiol 21: 954-956, 2000
- 18. Turjman F, Joly D, Monnet O, Faure C, Doyon D, Froment JC: MRI of intramedullary cavernous haemangiomas. Neuroradiology 37: 297-302, 1995
- 19. Zander DR, Lander P, Just N, Albrecht S, Mohr G: Magnetic resonance imaging features of a nerve root capillary hemangioma of the spinal cord: case report. Can Assoc Radiol J 49: 398-400, 1998