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Hemangiopericytoma of the Cervicothoracic Spine: A Case Report and Literature Review

Servikotorasik Omurga Hemanjiyoperisitomu: Bir Olgu Sunumu ve Literatür Derlemesi

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ABSTRACT

A case of hemangiopericytoma in the cervicothoracic spine is reported. A 43-year-old man was admitted to our hospital with pain in the scapular region radiating to the left upper extremity, numbness, and weakness of the legs. Magnetic resonance image revealed an intraspinal, extradural mass at the level of the C6-T2 vertebral bodies. The lesion also involved the dorsal paraspinal area with a giant mass. A total resection of the tumor was performed and histopathological findings revealed a malignant hemangiopericytoma. The patient showed rapid neurological function improvement after surgery. It is well-recognized that hemangiopericytoma is an aggressive tumor with a high risk of recurrence and propensity to metastasize. The tumor is rarely found in the central nervous system, and only a few reports could be found in the literature. We present a case of cericothoracic spinal hemangiopericytoma with an intensive review of the literature.

KEYWORDS: Hemangiopericytoma, Cervicothoracic spine, Extradural tumor, Total resection

ÖZ

Servikotorasik omurgada bir hemanjiyoperisitom olgusu sunulmaktadır. 43 yaşında bir erkek hastanemize skapüler bölgede oluşan ve sol üst ekstremiteye yayılan ağrı, uyuşukluk ve bacaklarda kuvvetsizlik ile geldi. Manyetik rezonans görüntüleme C6-T2 vertebral cisimler seviyesinde bir intraspinal, ekstradural kitle ortaya çıkardı. Lezyon ayrıca dev bir kitleyle dorsal paraspinal bölgeyi tutuyordu. Tümörün total rezeksiyonu yapıldı ve histopatolojik bulgular bir malign hemanjiyoperisitom gösterdi. Hasta cerrahiden sonra nörolojik olarak hızla iyileşti. Hemanjiyoperisitomun yüksek nüks riski ve metastaz eğilimi olan agresif bir tümör olduğu bilinmektedir. Tümör, merkez sinir sisteminde nadiren bulunur ve literatürde sadece birkaç olgu saptanmıştır. Literatürün kapsamlı bir gözden geçirilmesiyle bir servikotorasik spinal hemanjiyoperisitom olgusu sunuyoruz.

ANAHTAR SÖZCÜKLER: Hemanjiyoperisitom, Servikotorasik omurga, Ekstradural tümör, Total rezeksiyon

INTRODUCTION

Hemangiopericytomas (HPC) are rare vascular tumors that arise from pericytes, the contractile cells surrounding capillaries, also known as the pericytes of Zimmermann (47, 48). They were first described by Stout and Murray in 1942 (47). HPCs may occur anywhere in the human body but the most common locations are the extremities, pelvis, the retroperitoneum, and the head and neck areas (11). They are rarely found in the central nervous system and account for approximately 2% to 4% of all primary meningeal tumors (19,33). HPCs in the spine may involve the structures of the vertebra and the spinal canal, including the extradural and intradural space. Our search of the literature showed that 140 cases of HPC in the spine had been reported (1-17,19-46,49-56), and 64 cases were extradural (2, 4-6, 8-10, 15, 20-22, 24, 25, 27, 28, 31, 32, 34, 35, 37, 40-42, 44-46, 49, 51, 53-55) (Table I). Here we present a case of primary HPC invasion in the cervicothoracic spine, as well as presenting a review of the literature.

CASE REPORT

Clinical Presentation

A 43-year-old man was admitted to our hospital with pain in the scapular region radiating to the left upper extremity, numbness, and weakness of the legs. The intensity of the pain had progressed over a 5-year period before his admission. Three to four weeks before admission, he developed progressive weakness in the left upper extremity and lower extremities. He underwent acupuncture and took nonsteroidal anti-inflammatory medication without improvement. Physical examination revealed asymmetric paraparesis. Muscular atrophy was identified in the left upper extremity and legs. He had hyporeflexia and positive pathologic reflexes bilaterally. Bowel and bladder disorders were not observed.

Radiological Findings

Results of routine laboratory and cervical spine radiography were normal. Magnetic resonance (MR) imaging of the cervical spine was subsequently performed. The images

Table I: Summary of 65 Cases with Extradural Hemangiopericytoma in the Spine

Case No.	Series	Age (yrs)/sex	Level	Pain/ palsy	Treatment	Recurrence	Metastasis	Follow-up (mos)
1	Kumar R et al. 2007	16/F	T4-5	-/+	SR+RT	NA	NO	AWD (9)
2	Mohammadianpanah M et al. 2004	21/M	T2	+/+	SR+CT+RT	NA	NO	AWD (NA)
3	Akhaddar A et al. 2002	39/M	T4-6	+/+	TR+RT	NO	NO	CR (36)
4	Ciappetta P et al. 1985	36/F	C5-6	+/-	PR+RT	NA	NO	DOD (20)
5	Fathie K 1970	21/M	T6	+/+	TR	NA	NO	CR (17)
6	Harris DJ et al. 1978	28/M	C2-6	?/+	SR+RT	NO	NO	CR (56)
7		65/F	L2,	+/-	SR+RT	NO	NO	DOD (43)
8		46/M	C5	-/+	SR+RT	NA	YES	AWD (12)
9	Lin YJ et al. 1996	16/F	C2	+/-	TR	NO	NO	AWD (17)
10	Cole CD et al. 2009	36/F	C3	+/-	EM+TR+RT	NO	YES	AWD (48)
11	Santillan A et al. 2011	61/F	C2	+/+	EM+SR+RT	NA	NO	AWD (3)
12	Brass SD et al. 2004	53/M	T2-3	+/+	EM+SR+RT	YES	YES	AWD (36)
13	Stern MB et al. 1980	31/F	C6	+/+	TR	NA	NO	NED (12)
14	Woitzik J et al. 2003	48/F	C6-T2	+/+	TR+CT+RT	NO	YES	AWD (12)
15	Lee JK et al. 2006	55/F	C6-T1	+/+	SR+RT	YES	NO	AWD (8)
16	Nonaka M et al. 1998	49/F	T8	+/+	SR+RT	NA	YES	AWD (33)
17	Ijiri K et al. 2002	39/F	L1-2	+/+	TR	NO	NO	NED (24)
18	Chang CC et al. 2004	48/F	C2-3	+/+	EM+RT	NA	NO	AWD (12)
19	Radley MG et al. 1992	28/M	T7-8	+/+	TR+RT	NO	YES	AWD (16)
20	Scott M et al. 1974	38/M	T12-L1	+/-	SR+RT	YES	YES	AWD (228)
21	Cappabianca P et al. 1981	52/F	C6	+/+	ST+RT	NA	NO	DOD (1)
22	Muraszko KM et al. 1982	41/F	T12- L2	+/+	EM+TR	NA	NO	NED (≥14)
23		15/M	T11-L1	+/+	EM+TR+RT	NA	NO	NED (≥6)
24		11/F	T10	+/+	SR+RT	YES	NO	AWD (79)
25	Mao P et al. 1967	56/M	L3	+/-	No	NA	NO	DOD (NA)
26	Salvati M et al. 1991	29/F	L1-3	+/-	TR+RT	NO	NO	NED (120)
27	Cizmeli MO et al. 1992	20/M	L2	+/-	EM+TR+RT	NA	NO	NED (1.5)
28	Zentar A et al. 2009	42/F	S1-2	+/+	SR+RT	NA	NO	NED (5)
29	Tang JS et al. 1988	19/M	L2	+/+	TR+CT+RT	YES	NO	AWD (48)
30	Mena H, et al. 1991	47/M	T3-4	+/+	TR	NO	NO	NED (96)
31		58/M	T12-L1	+/-	SR+RT	YES	YES	DOD (36)
32	Hansen CP, et al. 1990	50/M	L3	+/-	RT	YES	YES	AWD (5)
33-51	Zhao Y, et al. 2007	NA	NA	NA	TR: 3 cases	YES: 11 cases	YES (4 cases)	NA
					TR+RT: 2 cases SR: 10 cases SR+RT: 4 cases	NO: 4 cases NA: 4 cases		
52	Yagishita A, et al. 1985	?/F	C7	+/+	TR	NA	NO	NA
53	Musacchio M, et al. 2003	56/F	C1	+/+	EM+SR+RT	YES	NO	AWD (48)
54	Schirger A et al. 1958	33/F	T2	+/+	SR	YES	NO	AWD (12)
55	Liu HG et al. 2013	31/F	C2-7	NA	SR+RT	YES	NO	AWD (19)
56		18/M	T11-L2	NA	TR+RT	YES	YES	AWD (65)
57		25/F	C2-3	NA	TR+RT	YES	NO	AWD (55)
58		37/M	C5-6	NA	TR+RT	YES	NO	AWD (72)
59		46/F	C1-4	NA	SR+RT	NO	NO	NED (25)
60		25/F	T12-L1	NA	SR+RT	YES	NO	AWD (34)
61		19/F	T5-6	NA	TR+RT	NO	NO	NED (30)
62		19/M	C7-T1	NA	TR+RT	YES	YES	AWD (54)
63		36/F	C1-2	NA	TR+RT	YES	NO	AWD (120)
64		14/M	L1-2	NA	SR+RT	YES	NO	AWD (24)
65	Present case	43/M	C6-T2	+/+	EM+TR	NO	NO	NED (12)

PR= *partial resection;* ***SR=*** *subtotal resection;* ***TR=*** *total resection;* ***CT=*** *chemotherapy;* ***RT=*** *radiotherapy;* ***EM=*** *embolization;* ***NA=*** *not applicable;* ***CR=*** *complete recovery;* ***AWD=*** *alive with disease;* ***DOD=*** *died of disease;* ***NED=*** *no evidence of disease.*

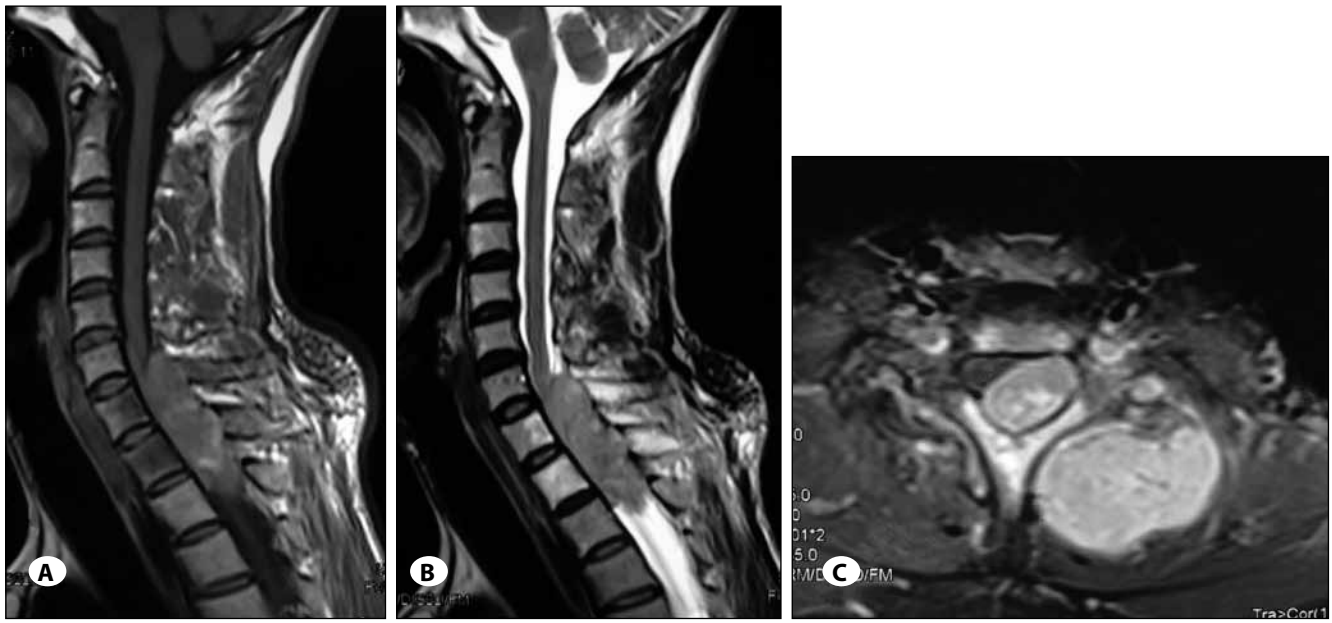


Figure 1: Sagittal magnetic resonance image showed extradural and paraspinal tumor at the C6-T2 level. It was isointense on both **A)** T1- and **B)** T2-weighted images. **C)** Axial T1-weighted magnetic resonance imaging after injection of gadolinium revealed a homogeneously enhancing extradural and paraspinal lesion at the cervicothoracic spine. The preoperative diagnosis was malignant schwannoma, neuroblastoma and neurofibroma.

revealed an intraspinal, extradural mass at C6-T2. The lesion also involved the dorsal paraspinal area with a giant mass. This mass was isointense on T1- and T2-weighted images, with homogeneous enhancement after administration of gadolinium (Figure 1A-C). Cystic components were also seen on gadolinium-enhanced MR images. The differential diagnosis made on the basis of preoperative imaging findings included malignant schwannoma, neuroblastoma and neurofibroma.

Management

Embolization was performed to reduce intraoperative blood loss before surgery. The patient underwent C6-T2 laminectomy and subsequently the spinal canal was opened. This revealed an intraspinal, extradural tumor posterolateral to the dura mater at this level. The tumor was adherent to the underlying dura. The tumor in the spinal canal was completely resected in a piecemeal manner. Then the mass in the dorsal paraspinal intramuscular spaces was excised. The tumor was hypervascular and hemorrhaging during tumor resection. In the end, we removed the tumor successfully. Posterior stabilization was achieved with pedicle screws and rods across C6 to T3.

The postoperative rehabilitation course was successful without complications or neurological deterioration. His upper and lower extremity motor functions improved gradually and, within 2 weeks after surgery the patient had recovered to ambulate with a neck collar. No disease progression or metastasis of the tumor was found during the 12-month follow-up period after surgery (Figure 2A,B). He is now well and closely followed-up.

Histopathology

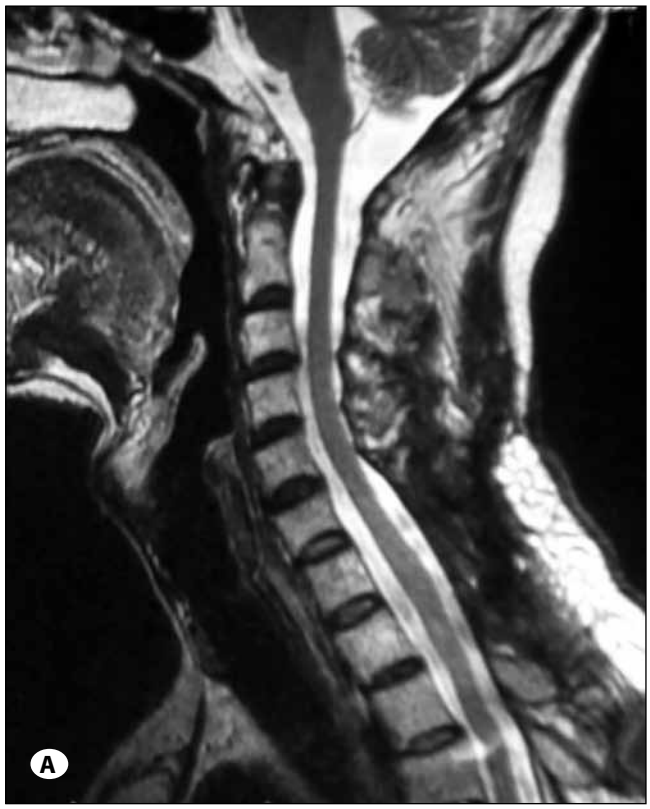
Microscopic examination demonstrated proliferation of tumor cells that showed round, oval or spindle shape nuclei with mitotic figures. The tumor was surrounded by abundant blood vessels with a sinus-like appearance that demonstrated a staghorn vasculature pattern typical of HPC (Figure 3). On immunohistochemistry, the tumor cells were positive for CD34, CD99, and Bcl-2, and negative for S-100 protein, CK, EMA, PR, CEA, and Desmin. This revealed that the pathologic type was anaplastic hemangiopericytoma, World Health Organization (WHO) grade III.

DISCUSSION

Hemangiopericytoma (HPC), a rare vascular tumor, is considered as an entity within the group of mesenchymal, nonmeningothelial tumors. The lack of large case series with long-term follow-up in the literature makes it difficult to reach a consensus on management of this disease.

HPC is a rare disease with non-specific imaging features and clinical manifestations making it difficult to reach a final or correct diagnosis on the basis of these presentations. Therefore, the diagnosis of this disease requires a combination of clinical and radiographic studies, and histological detection. Differential diagnosis of spinal HPC includes malignant schwannoma, neuroblastoma, neurofibroma, meningioma and solitary fibrous tumor (13, 14, 18, 19, 28, 56).

The initial and optimal management of HPC is total en bloc resection of the lesion (1-3, 6, 7, 22-24). It is well known that HPC has a propensity for recurrence and metastasis (4, 6, 13, 14, 19, 20, 25, 37, 45). Complete resection of the tumor



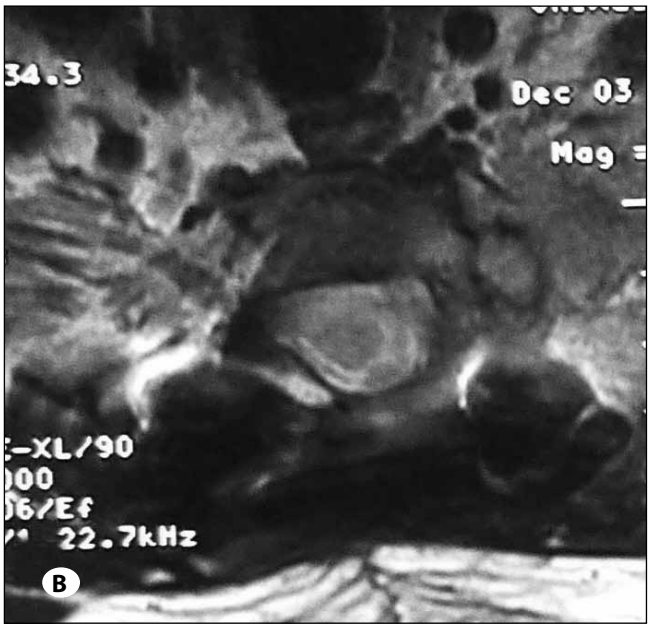


Figure 2: Postoperative T2-weighted magnetic resonance image scan in the **A)** sagittal and **B)** axial planes revealing decompression of the spinal cord and no indication of recurrence.

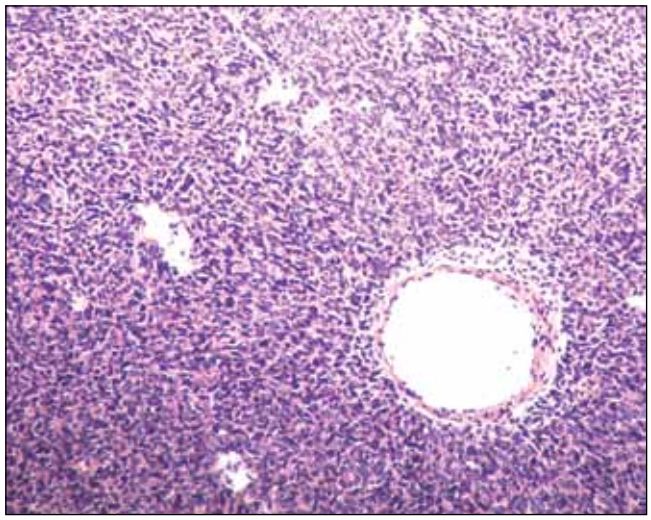


Figure 3: Hematoxylin and eosin staining (H&E; original magnification, ×100) showing large sinus-like and numerous dilated vascular branching in a staghorn appearance. Neoplastic cells exhibits diffusely around the vessels.

at the time of initial surgery should be pursued. However, HPC has marked vascularity and significant intraoperative bleeding, creating a great challenge for surgeons. This may lead to high surgery-related morbidity and mortality rates and a high risk of incomplete resection of the tumor. Preoperative endovascular embolization of tumors has been proven effective in controlling intraoperative blood loss and may facilitate surgical resection of the lesions (4, 6, 9, 10, 34, 35, 42). The present case received preoperative endovascular embolization. We encountered significant hemorrhage during the surgery but removed the tumor successfully. It is therefore important for the surgeons to carry out angiographic evaluation and preoperative embolization when they are suspicious of HPCs.

Despite the fact that surgery is the treatment of choice for HPC, it remains difficult to cure HPC with surgery alone. Some investigators have recommended that postoperative radiotherapy should be implemented to control the residual tumor (11, 13, 14, 19, 52, 55). In fact, adjuvant radiotherapy has been shown to be effective in controlling HPC (11, 13, 14, 19, 30, 55). However, the evidence of its efficacy has been mainly based on small retrospective studies. There was no statistically significant increase in survival among patients undergoing gross total resection plus adjuvant radiotherapy when compare with those undergoing gross total resection alone. In addition, as no clinical, randomized, prospective trials have been performed on this condition, there is no consensus on adjuvant radiotherapy as the routine treatment after surgery. In the presented case, no adjuvant radiotherapy was administered because of the lack of financial support and the aforementioned reason.

Most HPCs may relapse and/or metastasize over time after initial treatment (4, 20, 25, 35, 49, 55). The reported recurrence rate has ranged from 48% to 88%, with metastasis rates from 14% to 64% (14, 19, 22, 31, 33, 52). The rates of local recurrence and distant metastases are associated with the pathological grade of HPCs (19, 31, 33). Therefore, close long-term follow-up of HPC patients is necessary.

CONCLUSION

HPC has long been considered as an aggressive and lethal tumor. Total en bloc resection is the optional management for HPC, although intraoperative bleeding due to the high vascularity of this tumor may be the greatest hindrance. Preoperative embolization is recommended to facilitate surgical resection by reducing intraoperative blood loss. Close long-term follow-up is necessary because local recurrence and distant metastases can develop years after the initial treatment.

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