

# Capillary hemangioma of the cervical intervertebral disc

Ahmet Cetinkal · Ahmet Colak · Kivanc Topuz ·  
Cem Atabey · Ufuk Berber

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**Abstract** An anterior cervical decompression and fusion operation was complicated by extensive bleeding from the disc space. Histopathological evaluation of the resected specimen revealed the diagnosis as the very first reported case of capillary hemangioma in intervertebral disc space. Retrospective review of the preoperative MRI demonstrated supporting findings of a capillary hemangioma within the cervical intervertebral disc.

**Keywords** Capillary hemangioma · Cervical spine · Anterior cervical discectomy · Microsurgery · Intervertebral disc

## Introduction

Capillary hemangiomas (CHs) are generally considered to be malformations or hamartomas [1]. They are benign tumors or tumor-like lesions that originate from blood vessels and are most often encountered in the skin and soft

tissues, but have been rarely reported to develop in the brain or spinal cord [3, 5, 9, 10, 13–15, 17–20, 23]. The present case is the first reported case with a CH in the intervertebral disc space.

## Case report

A 76-year-old man was admitted with 3 years history of progressive neck, right shoulder and arm pain, partially relieved with the use of a collar. He also complained slight right-sided weakness and numbness, gait disturbance, tremor and urgency. Physical examination exhibited right dominant spastic quadriparesis and right-sided sensory deficit at C3, C4, C5 and C6 dermatomes.

Lateral cervical radiograph showed a clay shoveler's fracture of C5, but hence he had been an active rower sportsman up to his forties, this was reasonable (Fig. 1a). On magnetic resonance imaging (MRI), narrowing of the vertebral canal in C3–4/4–5/5–6 levels without signs of myelomalacia was evident (Fig. 2). He was planned to have an anterior C3–4 discectomy, C5 corpectomy and fusion operation (Fig. 1b).

Intraoperatively while incising the C4–5 disc, a surprising bleeding happened from the disc space. A vascular lesion was apparent with a closer look of the operation microscope magnification. The lesion was excised totally. Rest of the operation, corpectomy, discectomy and fusion were carried out uneventfully.

Since the lesion remained unrecognized until the time of operation, contrast enhanced MR images had not been obtained.

Histological evaluation of the specimen showed a vascular proliferation consisting of numerous capillaries lined by endothelial cells in disc matrix (Fig. 3).

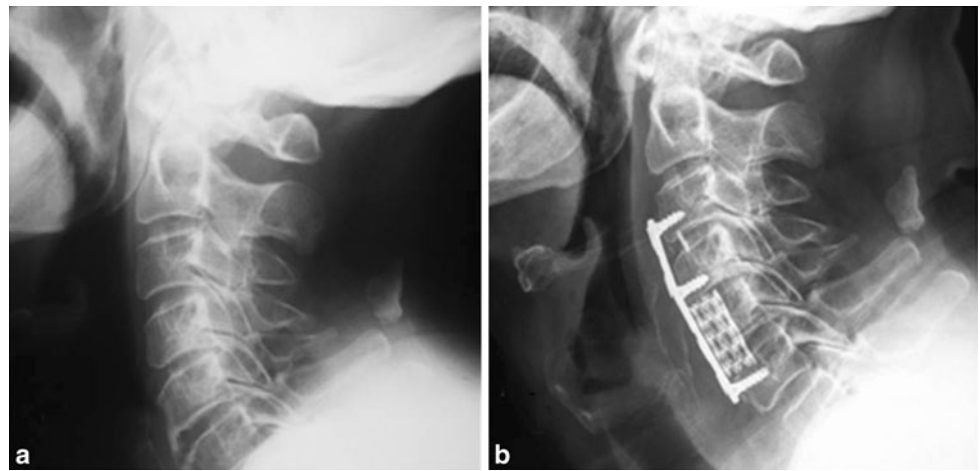
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A. Cetinkal (✉)  
Department of Neurosurgery, Kasimpasa Military Hospital,  
Kasimpasa, Beyoglu, Istanbul, Turkey  
e-mail: acetinkal@yahoo.com

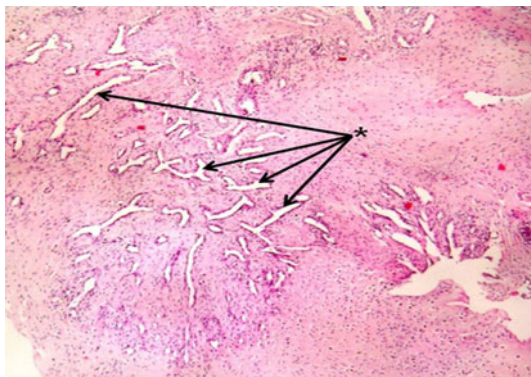
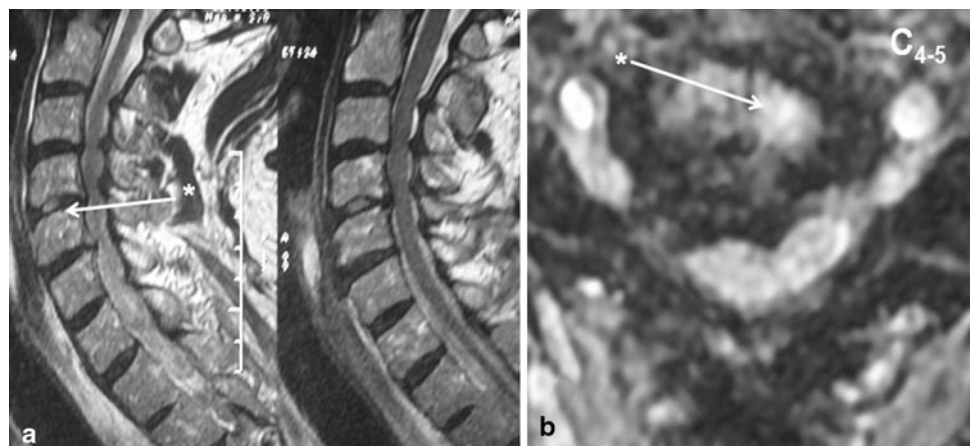
A. Colak · K. Topuz · C. Atabey  
Department of Neurosurgery,  
GATA Haydarpaşa Training Hospital,  
Uskudar, Istanbul, Turkey

U. Berber  
Department of Pathology, GATA Haydarpaşa Training Hospital,  
Uskudar, Istanbul, Turkey

**Fig. 1** **a** Preoperative sagittal X-ray (the spinous fracture of C5), **b** postoperative sagittal X-ray



**Fig. 2** **a** Sagittal T2WMRI (asterisk the heterogeneity in C4–5 intervertebral disc space. At the anterior part of C4–5 disc could be seen slightly hyperintense area), **b** axial T2WMRI of C4–5 (asterisk)



**Fig. 3** A pathological view of capillary hemangioma (Hematoxylin eosin,  $\times 40$ ) (asterisk vascular proliferation and numerous endothelial lined capillaries)

Hemosiderin pigment depositions were also seen in the lesion. Immunostaining with CD31,  $\alpha$ -smooth muscle actin and Ki-67 antigen were also performed. The final diagnosis was CH.

The patient's neurological condition improved within 18-month follow-up period.

## Discussion

CHs comprise the largest single group of this tumor type. Histologically, almost all lesions are composed of nodules of small, capillary-sized vessels, each of which is fed by an artery. The lobular or grouped arrangement of vessels is a very helpful feature in distinguishing CHs from malignant vascular proliferations. This group of lesions includes capillary hemangiomas of infancy, epithelioid hemangiomas, lobular capillary hemangiomas and variants of capillary hemangiomas [22]. Subtypes of CHs are well established before [1].

Usual locations of CH are soft tissues, cutaneous and subcutaneous tissues, and rarely osseous tissues, especially in spinal column [4, 20]. Vertebral hemangiomas are common pathologies, but those extending into the epidural space are rare [4, 6, 8, 11, 21]. Some purely epidural hemangioma cases reported are cavernous types [6, 8, 16]. The most frequent involvement site for CH in spinal cord and proximal nerves are intradural extramedullary compartment [12]. The occurrence of spinal epidural CH is exceedingly rare [21]. There are only four epidurally

located cases reported in the literature of CHs in the spinal channel [4, 7, 12, 21]. Following a wide search through English medical literature, we could not find any report about a CH located in the intervertebral disc space.

The pathogenesis of spinal CHs is not well established. They are thought to evolve from impaired migration and differentiation of primitive mesoderm at the embryonic mesodermal plate during the time when angioblastic differentiation starts [15]. It is implied that approximately one-third of the lobular CHs may develop following a minor trauma [1]. Since the presented case had a history of repetitive trauma due to rowing sports, the aforementioned hypothesis on pathophysiology may befit to this patient.

CHs were reported to be iso- or slightly hyperintense on T1-weighted, and hyperintense on T2-weighted MR images. Masses appeared to be homogeneous with strong enhancement following gadolinium injection [1, 21]. Sagittal and axial T2-weighted MR images of the present case showed heterogeneity and hyperintensity in C4–5 intervertebral disc (Fig. 2).

Histologically, CHs must be differed from cavernous hemangiomas, arteriovenous malformations and capillary telangiectasias. Unlike CHs, cavernous hemangiomas consist of dilated hyaline vessels and often exhibit thrombosis, perivascular hemosiderin deposition and calcification. Capillary telangiectasias and arteriovenous malformations of the central nervous system have neural parenchyma between vessels and lack lobularity and demarcation [2, 15].

CHs must also be distinguished from highly vascular neoplasms of the neuraxis, which include hemangioendotheliomas, hemangiopericytomas and hemangioblastomas [2, 15]. Hemangioendothelioma features cellular anomalies and an increasing number of cell mitoses. Pericellular deposition of reticulin fibers and collagen IV, as well as uniform spindle cells arranged around a central capillary are the characteristic findings for hemangiopericytoma. Unlike CH, hemangioblastoma contains foamy stromal cells and is not bordered by a fibrous pseudocapsule. Angiomatous meningioma that contains innumerable number of vessels is another diagnostic entity that must be differed from CH. Meningothelial cell nests are seated within the vascular channels and may be demonstrated by immunostains for epithelial membrane antigen [15].

Complete surgical removal is the treatment of choice for CH [15]. Because of the excessive vascularity of this lesion, piece to piece resection should be avoided to prevent bleeding in incomplete resection [20]. En bloc removal of the tumor should be preferred. Preoperative embolization of feeding arteries of the tumor with the selective spinal angiography may be another choice to deal with intraoperative bleeding [15]. Total discectomy and total excision of the lesion in the presented case provided successful result. Moreover, to distinguish the benign

nature of this tumor from a more malignant proliferation, preoperative imaging is not sufficient. Thus, immunohistological staining, including CD31,  $\alpha$ -smooth muscle actin and Ki-67 antigen, should also be accomplished.

As it is well-known, the intervertebral disc is avascular. Neither a CH nor any other lesion has been reported to exist in this region.

## Conclusion

Although rare, such a lesion can be recognized with special attention to MR images. Our case indicates the possibility of an unanticipated bleeding due to an unexpected vascular tumor in the disc space. En bloc surgical resection should be performed as surgical management of CH of the intervertebral disc to treat this tumor.

Usus est magister optimus! “Experience is the best teacher”.

**Conflict of interest statement** None of the authors has any potential conflict of interest.

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