Lumbar Intradural Extramedullary Capillary Hemangioma: A Case Report

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Abstract

Introduction: We report a case of intradural extramedullary (IDEM) capillary hemangioma.

Case report: A 59-year-old woman presented with a 2-month history of low back pain and paresthesia in the lower extremities. Magnetic resonance imaging (MRI) revealed a lobular tumor with an intense enhancement at the level of the L4 vertebral body. Tortuous tubular structures seen around the caudal pole of the tumor were considered to be a feeding vessel intermingled with nerve roots, which strongly suggested that the tumor was of a vascular origin. Operative findings demonstrated the dilated feeding vessel intermingled with the roots at the caudal pole of the mass. The mass was pathologically diagnosed as cavernous hemangioma.

Conclusion: A case of IDEM capillary hemangioma with MRI findings suggestive of a vascular tumor is reported. The MRI findings of the present case are different from those of capillary hemangiomas previously reported, and may be helpful in the differential diagnosis of capillary hemangioma from other IDEM tumors.

Keywords: Capillary hemangioma; Lumbar; Spinal; Intradural

Introduction

Capillary hemangiomas are benign vascular tumor or tumor-like lesions most often encountered in the skin or mucosa of the head and neck [1,2]. The occurrence of capillary hemangiomas within the confines of the spinal canal is rare [3]. Magnetic resonance imaging (MRI) findings of intradural extramedullary (IDEM) capillary hemangiomas are similar to those of other IDEM tumors, such as schwannomas and neurofibromas [4]. Therefore, they cannot be accurately differentiated from other IDEM tumors.

We report a case of a spinal IDEM capillary hemangioma. MRI revealed a lobular tumor with an intense enhancement at the level of the L4 vertebral body. Tortuous tubular structures seen around the caudal pole of the tumor on MRI were identified as the feeding vessel intermingled with nerve roots, which suggested that the tumor was of a vascular origin.

Case Report

A 59-year-old woman presented with a 2-month history of low back pain and paresthesia in the lower extremities. Pain and paresthesia occurred in the left lower extremity a month before the visit and the same symptom had developed more recently in the right lower extremity. These symptoms progressively worsened, and upon admission, she was unable to stand upright or walk due to the pain. Preoperative laboratory analysis and a neurological examination found no abnormalities.

MRI revealed an approximately 2-cm-sized well-defined lobular IDEM mass at the level of the L4 vertebral body. The mass was isointense with the spinal cord on T1-weighted images and hyperintense relative to the spinal cord on T2-weighted images. It showed homogenous strong enhancement on contrast-enhanced T1-weighted images. Conglomerates of tortuous tubular structures were seen around the caudal pole of the tumor (Figure 1A,B).
Figure 1: T2-weighted magnetic resonance (MR) images revealing an approximately 2-cm-sized well-defined lobular mass at the level of the L4 vertebral body (A, B). Conglomerates of tortuous tubular structures were seen around the caudal pole of the tumor (white arrow). On contrast-enhanced T1-weighted sagittal (C) and axial (D) MR images, the structures showing tortuous and nodular enhancement with multiple void signal intensity (black and white arrows). MR myelography showing a total blockage of the cerebrospinal fluid by the mass, with prominent venous engorgement from the mass up to the spinal cord (E, black and white arrows).

The structures were isointense on T1-weighted images and hypointense on T2-weighted images. On contrast-enhanced T1-weighted images, the structures showed tortuous and nodular enhancement with multiple void signal intensity (Figure 1C,D). Magnetic resonance (MR) myelography showed a total blockage of the cerebrospinal fluid by the IDEM mass. It also demonstrated prominent venous engorgement from the mass up to the spinal cord (Figure 1E).

We performed a spinous process splitting laminectomy between the L3 and L4 levels [5]. Following the incision at the dural midline, a dark reddish mass adherent to a single rootlet was identified. There was no evidence of acute or chronic hemorrhage around the mass. A dilated feeding vessel intermingled with the roots was identified at the caudal pole of the mass and was resected after coagulation. Next, the mass was carefully dissected from the surrounding roots and completely removed. After surgery, the patient reported significant improvement of her back and leg pain as well as the paresthesia. Upon histological examination, the lesion showed a lobular proliferation of well canalized and poorly canalized vessels. The vessels were lined with plump endothelial cells with small lumens. This finding was consistent with a capillary hemangioma (Figure 2).

**Discussion**

The majority of capillary hemangiomas are superficial lesions found in the skin or mucosa of the scalp, face, nasal vault, oral cavity, and neck [2]. They have rarely been encountered in the neuroaxis [6]. So far, more than 30 cases of spinal intradural capillary hemangioma have been reported, including the present case [7,8]. Spinal intradural capillary hemangiomas commonly present in the fifth and the sixth decades of life, with no obvious gender predilection [8]. They usually present at the surface of the distal spinal cord, close to the conus medullaris or attached to the nerve roots of the cauda equine [3].

Capillary hemangioma is a benign vascular tumor or tumor-like lesion, which 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 [8]. However, most capillary hemangiomas studied did not show typical MRI findings suggestive of a vascular tumor, such as a vascular flow void corresponding to feeding arteries and draining veins or marked venous engorgement [1-4,6,8,9]. In the present case, the capillary hemangioma was isointense with the spinal cord on T1-weighted images and hyperintense relative to the spinal cord on T2-weighted images. The mass showed homogenous strong enhancement on contrast-enhanced T1-weighted images. These MRI findings correspond well with those of previous reports [1-3,6,9,10]. Alternatively, capillary hemangiomas have also been reported to be hyperintense on T1-weighted images, isointense on T1-weighted images and hypointense on T2-weighted images, or isointense on both T1- and T2-weighted images [4,8,11]. Considering these heterogeneous MRI findings, it is usually difficult to accurately differentiate capillary hemangiomas from other IDEM tumors, such as schwannomas, and neurofibromas [4,10]. Schwannomas and neurofibromas usually appear hyperintense on T1- and T2-weighted images with homogenous strong enhancement on contrast-enhanced T1-weighted images. However, sometimes they may be relatively isointense on both sequences [4].

Most spinal intradural capillary hemangiomas did not show MRI findings suggestive of a vascular tumor [1-4,6,8-11]. Zander et al. proposed that extramedullary arteriovenous malformations may be differentiated from capillary hemangiomas on MRI by a vascular flow void corresponding to feeding arteries and draining veins [4]. However, the present case showed MRI findings suggestive of a vascular tumor. Conglomerates of tortuous tubular structures were seen around the caudal pole of the tumor.
The structures were isointense on T1-weighted images and hypointense on T2-weighted images. There were multiple void signal intensities on contrast-enhanced T1-weighted images. Operative findings demonstrated these structures to comprise a feeding artery intermingled with nerve roots. The identification of draining veins on MRI may be helpful in differential diagnosis of vascular tumors such as hemangioma, hemangioblastoma, and paraganglioma from nonvascular tumors [8,12]. MR myelography of the present case showed prominent venous engorgement from the mass up to the spinal cord, which also suggested that the tumor was of a vascular origin.

In the present case, we removed the tumor using spinous process splitting laminectomy between L3 and L4. Spinal process splitting laminectomy, introduced by Watanabe et al. [13], is one of the alternative surgical technique for conventional laminectomy, preserving posterior supporting structures [5]. Although spinous process splitting laminectomy has usually been performed for the treatment of lumbar spinal stenosis, the procedure is also useful in spinal tumor surgery. Since it allows wide exposure of the tumor, effective removal of tumor is possible while preserving posterior supporting structures.

Conclusion

A case of IDEM capillary hemangioma with MRI findings suggestive of a vascular tumor is reported. The MRI findings of the present case are different from those of capillary hemangiomas previously reported, and may be helpful in the differential diagnosis of capillary hemangioma from other IDEM tumors.

References


Figure 2: Histological view of the lesion demonstrating lobular proliferation of well canalized and poorly canalized vessels. The vessels are lined by plump endothelial cells with small lumens (hematoxylin and eosin staining, original magnification ×400).
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