

# Pure Epidural Cavernous Hemangioma of the Cervical Spine that Presented with an Acute Sensory Deficit Caused by Hemorrhage

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Pure epidural cavernous hemangioma of the spine without vertebral involvement is rare. Due to the slow growth of this lesion, the most common symptoms are chronic pain, myelopathy, and radiculopathy. In our case, the patient complained of an acute onset sensory deficit of the C4 dermatome. An MRI revealed an epidural mass with an acute hematoma. Here, we report a case of a pure epidural cavernous hemangioma that presented with acute neurologic symptoms caused by intralesional hemorrhage and an acute epidural hematoma, which were demonstrated on the patient's MRI.

**Key Words:** Cavernous hemangioma, spine, magnetic resonance imaging, epidural space

## INTRODUCTION

Cavernous hemangiomas are vascular malformations that appear frequently in the intracranial structures, but are rare at the spinal level. Most epidural hemangiomas are secondary extensions from vertebral lesions, and pure epidural hemangiomas are rare.<sup>1-4</sup>

We report the case of a pure epidural hemangioma that presented with acute pain and hyper-

esthesia of the C4 dermatome. Radiological findings showed an acute hemorrhage in the lesion and epidural space.

## CASE REPORT

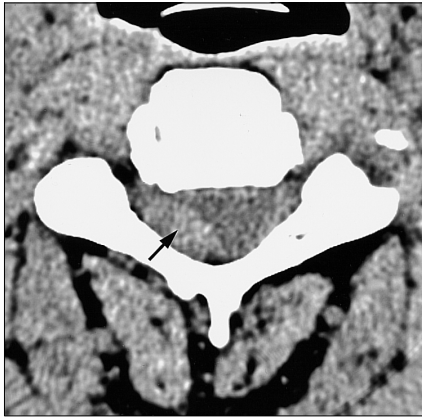
A 48-year-old man was admitted with acute onset pain and hyperesthesia of the right lower neck and shoulder that had lasted for five days. His symptoms were worse at night. He had no history of hypertension or trauma and no current use of medications. The neurological examination revealed normal motor strength and normal deep tendon reflexes of the upper and lower extremities. Sensation to the pinprick test was increased over the right C4 dermatome, including the right side of the lower neck and right shoulder. Laboratory findings were unremarkable with no evidence of coagulopathy. CT scan revealed an oval-shaped, slightly hyperdense mass displacing the thecal sac to left (Fig. 1. black arrow). T2-weighted sagittal MR images revealed a 1.5 × 0.7 centimeter sized, elliptical lesion posteriorly-located on the right side of the C3-4 epidural space. The spinal cord was displaced anteriorly and a thin, hypodense band was noted between the lesion and the spinal cord (Fig. 2A). The thin layer of low signal intensity represented the dura and indicated that the tumor was extradural. This lesion was isodense with the spinal cord on a T1-

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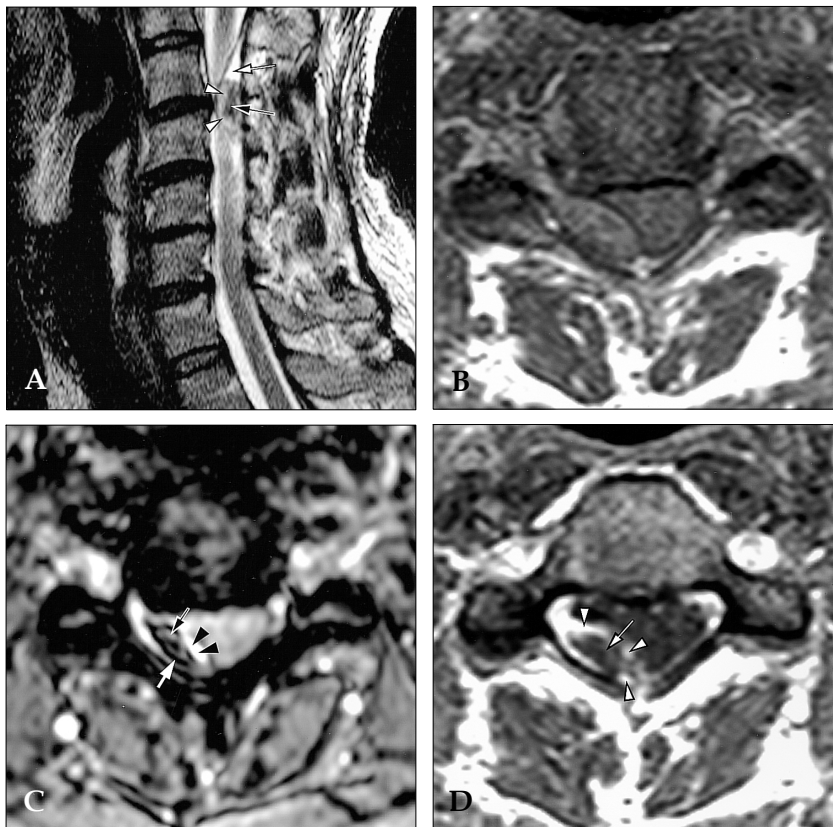
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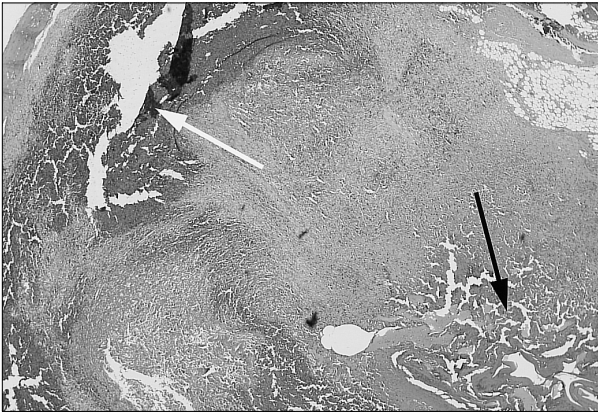


**Fig. 1.** Axial CT image shows slightly hyperdense, oval-shaped mass (arrow) anterior to the right lamina.

weighted axial image (Fig. 2B). The axial gradient-echo showed an oval-shaped epidural mass with a peripheral dark signal rim and a central hypodense area (Fig. 2C, white arrow). The remaining portion of the mass showed bright signal intensity on a gradient-echo image (Fig. 2C, black arrow) and on a T2-weighted sagittal image (Fig. 2A, white arrowhead). After contrast enhancement, the peripheral portion of the lesion enhanced slightly (Fig. 2D, white arrow) but the central portion showed no enhancement. We assumed that this dark signal area on the gradient-echo and T2-weighted images was an acute hematoma containing deoxyhemoglobin or hemosiderin deposi-



**Fig. 2.** (A) Sagittal T2-weighted image shows a posterior epidural mass with peripheral high signal intensity (white arrowheads) and a central low signal area (black arrow). The mass is well-demarcated from the spinal cord by a dark signal line representing the dura. The triangular shaped epidural lesion above and below the main lesion shows high signal intensity (white arrow). This area was later confirmed as a hyperacute hematoma in the epidural space. (B) This lesion was isodense with spinal cord on axial T1-weighted image. (C) Axial gradient-echo (fast field gradient echo, TR/TE/FA, 536/23/25 degree) image shows very dark central signal area and a peripheral dark signal rim similar to the area in the peripheral portion of the mass (white arrow). Initially, this area was thought to be a hemosiderin rim; however, pathological findings showed it be an acute hematoma. This dark signal characteristic of the gradient-echo image may be caused by deoxy-hemoglobin in the hematoma. The remaining mass shows high signal intensity (black arrow). Outside of the main lesion is a bright signal epidural lesion (black arrowheads) that was confirmed to be a hyperacute hematoma in the epidural space. (D) After Gadolinium-DTPA contrast enhancement, the mass shows peripheral heterogeneous enhancement (white arrow). Surrounding epidural hyperacute hematoma shows strong contrast enhancement (white arrowheads).



**Fig. 3.** Photomicrograph shows a cavernous hemangioma composed of thin-walled vascular channels in collagenous tissue. There was a large central hematoma (black arrow) and peripheral hematoma (white arrow) (H&E stain,  $\times 40$ ). There was no hemosiderin deposition in the mass.

tions from the repeated hemorrhage of the mass. There was also an epidural lesion surrounding the main lesion which was hyperdense on T2-weighted (Fig. 2A, white arrow) and gradient-echo images (Fig. 2C, black arrowhead). This lesion was isodense on T1-weighted images and showed strong contrast enhancement after Gd-DTPA infusion (Fig. 2D, white arrowhead). The enhancing epidural lesion outside of the main lesion that showed iso-signal intensity on the T1-weighted image and high signal intensity on the T2-weighted and gradient-echo images suggested a hyperacute hematoma in the epidural space.

We performed a hemi-laminectomy and total extirpation of the mass. Upon visual inspection the mass appeared to be a purple colored, well-vascularized mass with marginal and central hematomas and was localized to the epidural space. An acute epidural hematoma surrounded the lesion. Pathologically, the lesion consisted of a large number of vascular channels in collagenous tissue, mainly of the cavernous type, and there were hematomas in the central (white arrow) and peripheral portions (black arrow) of the lesion (Fig. 3). There was no hemosiderin deposition in the mass. The pathological diagnosis was cavernous hemangioma. Postoperatively, the patient's symptoms resolved and his recovery from the surgery was uneventful. At 1-year follow-up examination, he remained free from the

pain he had experienced preoperatively.

## DISCUSSION

Cavernous hemangiomas are vascular malformations that constitute 5-12% of spinal vascular lesions. Pure epidural involvement is rare and is usually the extension of a vertebral lesion into the epidural space. Pure epidural hemangiomas of the spine without bone involvement are very rare and they are most often located in thoracolumbar areas; fewer than 10 cases of cavernous hemangiomas at the cervical level have been reported.<sup>1-4</sup> The onset of symptoms may be acute, progressive, or remittent, depending on the biological behavior of the tumor. Acute symptoms are rare and generally due to intralesional hemorrhage or thrombosis.<sup>5</sup>

Most pure epidural hemangiomas are isodense compared with those of the spinal cord on T1-weighted images. On T2-weighted images, the signal intensity of the lesions is frequently high. After contrast enhancement, these lesions show strong, homogeneous enhancement. Intraaxially located cavernous hemangiomas frequently show a peripheral low signal intensity rim, representing hemosiderin deposition from recurrent intralesional hemorrhage. Epidural cavernous hemangiomas of the spine usually do not show a low signal rim. This is presumably caused by the easier removal of blood products outside the blood-brain barrier.<sup>6-9</sup> In our case, there was a low signal rim detected in the peripheral portion of the mass on a gradient-echo axial image. Pathological reports showed an acute hematoma in the peripheral portion of the mass with no hemosiderin deposition. Gradient-echo images are very sensitive to small, hemorrhagic products, such as deoxyhemoglobin, methemoglobin, and hemosiderin. Pathologic findings showed the low signal rim in the peripheral portion of the mass on the gradient-echo image to be an acute hematoma containing deoxyhemoglobin.

This case had several unusual clinical and radiological features. First, to our knowledge, our patient's lesion was located at the highest level to be reported in the literature about spinal epidural hemangiomas. Second, based on the MRI findings,

we were able to illustrate, in detail, the intraleisional acute hematoma that caused the patient's symptoms. There was also a hyperacute epidural hematoma surrounding the main lesion. This hyperacute hematoma showed typical MR findings including isodensity on the T1-weighted image, hyperdensity on the T2-weighted image, and strong contrast enhancement on T1-contrast enhanced image., The hyperacute epidural hematoma may have been another cause of the patient's acute symptoms. Spontaneous spinal epidural hemorrhage is rare and accounts for less than 1% of all spinal epidural lesions.<sup>10</sup> The most common causes include coagulopathies, hypertension, increased venous pressure and vascular malformations. Kubo et al.<sup>11</sup> studied 99 cases of spinal epidural hematoma and in 14 (14%) of these cases, pathological examination revealed vascular malformations. Graziani et al.<sup>12</sup> reported similar results, which showed vascular malformations in 18% of the patients they studied. Spontaneous epidural hematomas are often thought to be venous in origin. Since the venous pressure in the cervical epidural veins is less than the intrathecal pressure, the possibility of severe spinal cord compression by this type of bleeding is low. Therefore, arterial bleeding is considered to be a more likely source of spontaneous spinal epidural hematoma. Urgent decompression surgery has been the primary treatment for spinal epidural. However, there have been several reports of spontaneous resolution of spinal epidural hematoma without surgery. Nonsurgical treatment is recommended for rapid neurological deterioration that is followed by early clinical recovery with and in cases where radiological studies confirm resolution of the lesion.<sup>13,14</sup>

The lesion should be differentiated from other epidural, neoplastic or inflammatory conditions, such as meningioma, lymphoma, metastases, hemorrhagic vascular mass, and epidural abscess. In our case, there was no fever or inflammatory signs suggesting epidural abscess, no demonstrable primary malignant focus, no hematologic abnormalities, and no vascular signal voids suggestive of arteriovenous malformations. We were able to diagnose the hemorrhagic vascular tumor by MRI findings.

In summary, we report a case of pure epidural

cavernous hemanigoma manifesting with intraleisional and acute epidural hemorrhage. Although rare, proper surgical management may be warranted.

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