Spinal epidural vascular malformation presenting in association with a spontaneously resolved acute epidural hematoma

Case report

YASUSHI MIYAGI, M.D., PH.D., MASAYUKI MIYAZONO, M.D., PH.D., AND KAZUFUMI KAMIKASEDA, M.D., PH.D.

Department of Neurosurgery, Kaizuka Hospital, Fukuoka, Japan

A 16-year-old boy developed acute neck pain and severe quadriparesis after mild rotatory movement of his neck. Magnetic resonance imaging revealed a cervical epidural hematoma that resolved spontaneously within a few days. Vertebral angiography demonstrated a small vascular malformation in the upper cervical epidural space. The vascular mass on the dural surface was totally resected and confirmed to be an arteriovenous malformation. This case represents the importance of including routine angiography in designing therapeutic strategy for cases of spinal epidural hematoma with spontaneous resolution.

**KEY WORDS** • spinal epidural hematoma • epidural vascular malformation • spine

A 16-year-old boy developed acute neck pain and severe quadriparesis after mild rotatory movement of his neck. Magnetic resonance imaging revealed a cervical epidural hematoma that resolved spontaneously within a few days. Vertebral angiography demonstrated a small vascular malformation in the upper cervical epidural space. The vascular mass on the dural surface was totally resected and confirmed to be an arteriovenous malformation. This case represents the importance of including routine angiography in designing therapeutic strategy for cases of spinal epidural hematoma with spontaneous resolution.

**Case Report**

**History.** This 16-year-old boy developed sudden and severe neck pain followed by complete quadriparesis and hypesthesia below both shoulders within a 30-minute period. The patient was admitted to our hospital by ambulance.

**Examination.** At admission, the patient was alert and his weakness had already begun to subside. The deep tendon reflexes of his extremities were hyperactive, and bilateral positive Babinski’s signs were present.

Emergency MR imaging of the cervical spine revealed an epidural hematoma extending from C-2 to C-6 (Fig. 1A). The lesion appeared as a high-intensity signal on both T1- and T2-weighted MR images. The hematoma was thickest at C-3, compressing the cervical spinal cord anteriorly. Because the patient’s neurological symptoms continued to improve after admission, he was carefully observed while preparations for emergency surgery were made. The day after admission, both the quadriparesis and sensory disturbance disappeared completely. On MR images, the hematoma had greatly decreased in size; the massive effect on the cervical spinal cord had also diminished and finally disappeared 2 weeks after admission (Fig. 1B and C). After the hematoma disappeared, intravenous administration of gadolinium–diethylenetriamine pentaacetic acid revealed no abnormal contrast enhancement. A vertebral angiogram, however, revealed small abnormal contrast pooling in the dorsal epidural space between C-1 and C-2 from the early arterial phase to the venous phase (Fig. 2). Because the lesion was diagnosed as being a hemorrhagic epidural vascular malformation, an operation was performed to prevent possible rerupture of the vascular malformation.

**Operation and Pathological Findings.** The patient underwent a laminotomy at C-2. Under the arch of C-2, an angioma-like vascular mass consisting of fine vasculature was found on the dorsal surface of the cervical dura mater.
A small draining vein ran anterolaterally on the left side of the dural surface. The mass had no vascular connection to the spinal cord and did not penetrate the dura mater. The lesion was totally resected from the dural surface. Histopathological analysis of the mass revealed an arteriovenous malformation (Fig. 3).

Postoperative Course. The patient made an uneventful recovery and was discharged without any neurological deficits 1 month after surgery.

Discussion

Some authors believe “spontaneous SEH” includes all cases in which trauma is not the cause, whereas others define spontaneous SEH as including only cases with an unidentified cause. In either case, this entity includes multifactorial bleeding under possible but unidentified basic hemorrhagic conditions, such as anticoagulation therapy, pregnancy, and blood dyscrasia.

Spontaneous SEH is a rare condition, normally seen in the lower cervical and dorsal regions in children and adolescents and in the thoracic or thoracolumbar regions in adults. In males, spontaneous SEH occurs most frequently in the lower cervical region, whereas in females the most common site is the lower thoracic region. Hematomas in the upper cervical spine (such as in our case) and in the lumbar and sacral area are very rare. It has been suggested that congestion followed by rupture of the spinal venous plexus is the primary event. However, venous pressure in the cervical epidural veins is less than intrathecal pressure at the same level, and hemorrhage from an epidural vein would not significantly compress the spinal cord. Therefore, arterial bleeding is currently considered to be the main symptom of acute spinal cord compression.

Although computerized tomography (CT) scanning demonstrates SEH as a high-density area in the epidural space with displacement of the spinal cord to the opposite side. The following figures illustrate the chronological series of sagittal T₁-weighted MR images showing an epidural hematoma located posterior to the spinal cord and extending from C-2 to C-6. Images were obtained at onset of symptoms (A), the next day (B), and 2 weeks later (C).
Spontaneous spinal epidural hematoma

side, this type of imaging sometimes is nondiagnostic. In the case of acute onset of neck pain associated with radiculopathy or myelopathy, emergency MR imaging should be performed even if there is no history of neck trauma. The sagittal MR images clearly reveal the appearance of epidural hematoma protruding into the spinal canal and demonstrate the precise extension of the hematoma. However, great care must be taken in diagnosing the origin of the hemorrhage because SEH does not always appear around the hemorrhagic focus. In fact, the vascular malformation in our case was located at C-2, whereas the SEH was located between C-2 and C-6. It is, therefore, suggested that the SEH easily extended caudally in the spinal epidural space where the connection of yellow ligaments and the dura mater are not tight. Acute SEH appears as a signal that is isointense with that of the spinal cord on T1-weighted images and as a heterogeneous signal on T2-weighted images; on the other hand, subacute SEH displays a high-intensity signal on both T1- and T2-weighted images.22 In our case, there may have also been multiple hemorrhagic events, because the MR images demonstrated typical signal patterns of subacute hemorrhage.

Some physicians believe that venous bleeding leads to the formation of spontaneous SEH; however, others have also found some vascular malformations among nontraumatic SEHs.6,13 Angiography is not routinely performed in the diagnosis of SEH without any significant findings on MR imaging.10 and angiographic demonstration of epidural vascular malformations is very rare.14 Kubo, et al.,9 studied 99 cases of SEH; in 14 (14%) of these cases, pathological examination revealed vascular malformations. Graziani and colleagues7 observed two (18%) of 11 SEH cases in which there were vascular malformations of the spinal epidural space. Olivero and associates10 reported cases in which vascular malformations in the epidural space were demonstrated angiographically but were not suspected on MR imaging. In our case, the MR images indicated a hematoma in the subacute stage, whereas the symptoms suggested an acute phase. These findings may be explained by possible small repetitive hemorrhagic events that developed after an acute catastrophic neurological deterioration. Therefore, a routine check of such vascular lesions by angiography seems to be very important in cases in which there is spontaneous SEH.

It is accepted that a laminectomy must be performed immediately after radiological confirmation of spontaneous SEH to prevent further progression of the neurological deficits.16 On the other hand, some cases resolved spontaneously with conservative treatment.2,4,8 Nonsurgical treatment is recommended for rapid neurological deterioration that is followed by an early and sustained neurological recovery, as confirmed by radiological resolution of the lesion.4 In cases of spontaneous SEH, risk factors for rebleeding and recurrence of SEH have not yet been elucidated,8 as opposed to cases of cranial epidural hematomas. Therefore, in our opinion, an angiogram should be routinely obtained, even in cases in which there has been spontaneous resolution of an SEH. In addition, these findings suggest that an acute onset of symptoms with a subacute MR pattern may indicate the existence of a repetitive source of hemorrhage, such as a vascular malformation.

References


Manuscript received September 30, 1997.
Address reprint requests to: Yasushi Miyagi, M.D., Ph.D., Department of Neurosurgery, Kaizuka Hospital, 7-7-27 Hakoizaki, Higashi-Ku, Fukuoka 812, Japan.

J. Neurosurg. / Volume 88 / May, 1998

911