Spontaneous spinal epidural hematoma inducing acute anterior spinal cord syndrome

Case report

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Spontaneous spinal epidural hematoma (SSEH) is rare. Its etiology remains controversial; however, spinal venous wall susceptibility to intravenous pressure change and the resultant venous rupture seem to be involved. The authors report a case of SSEH dorsal to the spine producing acute anterior spinal cord syndrome. A posterior SSEH between the C-3 and T-5 levels caused progressive tetraparesis and the disappearance of superficial body sensation below the level of C-8, although deep sensation remained completely intact. This neurological false localizing sign seems to have resulted from counterforce by preexisting asymptomatic cervical intervertebral disc herniation at the C6–7 levels inducing direct pressure on the anterior spinal cord. This case is the first reported instance of posterior cervical SSEH manifesting acute anterior spinal cord syndrome as its false localizing sign. (DOI: 10.3171/2009.2.SPINE08342)

Key Words • spontaneous spinal epidural hematoma • false localizing sign • acute anterior spinal cord syndrome

Spontaneous spinal epidural hematoma is rare. Its etiology remains unclear,14 but early studies have suggested that spinal venous susceptibility is an important causative factor.5–8 The cord compression caused by SSEH results in the transverse disruption of the spinal neuronal tracts and induces sensorimotor dysfunction below the level of the spinal injury.7 We report on a case of posterior cervical SSEH that caused anterior spinal cord syndrome. We describe the clinical profile and discuss the possible mechanism of the neurological false localizing sign for the posterior SSEH.

Case Report

History and Examination. This 63-year-old previously healthy woman experienced a sudden severe pain in the back of her head and neck while sitting calmly. Subsequently, she suffered from progressive weakness of the right lower extremity and was immediately transferred to our hospital with normal vital signs and clear consciousness. The neurological examination on admission revealed complete paralysis of the bilateral lower extremities, paresis of both hands, and the symmetrical disappearance of superficial body sensation (pain and temperature) below the spinal level of C-8. Note, however, that deep sensation (vibratory and joint sensations or proprioception) and the fine touch sense remained intact throughout the study period. The deep tendon reflexes disappeared bilaterally in the patellar and ankle joints. Blood examinations showed normal results including those for the coagulatory and fibrinolytic systems and hemostatic function.

Neuroimaging Findings. An MR image obtained on admission showed an acute epidural hematoma located at the right dorsal aspect of the spine between the C-3 and T-5 levels (Fig. 1A and B). The hematoma had its maximum diameter at C-7. The MR image also revealed a central-type CIDH at the C6–7 level that had been asymptomatic. Therefore, the spinal cord was markedly compressed at C6–7 (right > left) by the CIDH and hematoma. Gadolinium-enhanced T1-weighted MR imaging revealed no change suggestive of tumor, inflammation, or vascular malformation.

Operation. The patient underwent surgical removal of the epidural hematoma as well as spinal cord decompression with a C6–7 laminectomy 7 hours after symptom onset. Intraoperative exploration showed no abnormal vascular lesion or malformation or neoplasm. The posterior SSEH was diagnosed based on MR imaging, intraoperative findings, and histological study.

Abbreviations used in this paper: CIDH = cervical intervertebral disc herniation; IVVP = internal vertebral venous plexus; SSEH = spontaneous spinal epidural hematoma.
Anterior cord syndrome in spontaneous spinal epidural hematoma

Postoperative Course. Postoperative MR imaging showed successful decompression of the spinal cord; however, the T2-weighted MR image revealed a spinal parenchymal change of hyperintensity (Fig. IC and D). The patient completely recovered from the neurological deficits after surgery, except for slight motor clumsiness of the right C-8 distribution, which persisted 6 months after treatment. Follow-up MR imaging performed 6 months after the onset of symptoms demonstrated a small cystic change in the spinal gray matter of the C-8 segment without any spinal compression (Fig. 1E and F).

Discussion

An SSEH is rare. Authors of an early study have described its incidence as 0.1 case per 100,000 population per year. Spontaneous spinal epidural hematomas primarily develop in persons between 50 and 80 years of age (male/female ratio 1.4:1) and predominantly occur dorsal to the spine at either the cervicothoracic or thoracolumbar levels. They are defined as spinal epidural hematomas developing without any specific cause including trauma, vascular malformation, neoplasm, or systemic hemorrhagic tendency. Table 1 lists the early reports of SSEHs in adults (≥ 18 years old) in which MR imaging reveals evidence of a spinal epidural hematoma and patient activity at the onset of neurological deficits is clearly described. The bleeding mechanism of an SSEH remains unclear. Early studies, however, have suggested that an SSEH results from a vulnerability of the spinal venous system and less likely from a susceptibility of the arterial component. Groen and colleagues have studied the etiology of SSEHs by investigating clinical cases and human cadavers, and based on the anatomical and developmental characteristics of the spinal venous system, they concluded that the posterior IVVP plays an important causative role in SSEH. The valveless, thin-walled venous plexus seems at risk for rupture caused by venous pressure changes. The fact that most SSEHs develop dorsal to the spinal cord also supports the theory that the posterior IVVP is the main bleeding source of these hematomas. On the other hand, some authors have pointed out the significance of epidural arterial rupture in SSEH. We were unable to identify the particular cause of the SSEH in our patient in terms of the venous or arterial origin; however, the location of the SSEH was dorsolateral to the spinal cord. This finding lends support to the contention that the posterior IVVP is the bleeding source for an SSEH.

Fig. 1. Sagittal (A) and axial (B) T2-weighted MR images of the cervical spine obtained 4 hours after the onset of neurological deficits, showing an epidural hematoma in the spinal canal posterior to the spine. The hematoma size is maximal at the C-7 level. The intervertebral disc herniation at C6–7 is significant (A). The axial view (B) at the C-7 vertebral body level shows the hematoma to the right of, and posterior to, the spinal cord. The hematoma compresses and pushes the spinal cord in a left ventral direction. Sagittal (C) and axial (D) spinal T2-weighted MR images obtained 4 days after hematoma removal, confirming successful spinal decompression. A hyperintense change (C) is apparent in the spinal cord centered at the C6–7 level. The axial image (D) at the C-7 vertebral body level shows hyperintensity, indicating edema in the right spinal gray matter. Sagittal (E) and axial (F) spinal T2-weighted MR images obtained 6 months after the onset of neurological deficits, demonstrating a small localized hyperintensity area suggestive of necrotizing cystic change in a portion of the right anterior horn. No spinal cord compression remains.
<table>
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<th>Authors &amp; Year</th>
<th>Age (yrs), Sex</th>
<th>Activity at Onset</th>
<th>Symptom at Onset</th>
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<th>Hematoma Location in Spinal Canal on MRI</th>
<th>Neurological Findings on Admission</th>
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<th>Outcome (FU period)</th>
<th>FU MRI (duration from onset to MRI)</th>
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<tr>
<td>Serizawa et al., 1995</td>
<td>46, F</td>
<td>bending forward</td>
<td>back pain</td>
<td>T3–6</td>
<td>pst</td>
<td>normal</td>
<td>conservative</td>
<td>complete recovery (1 wk)</td>
<td>hematoma resolved (30 days)</td>
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<td>Jamjoom, 1996</td>
<td>79, M</td>
<td>lifting heavy object</td>
<td>middorsal pain</td>
<td>T6–8</td>
<td>pst</td>
<td>paresis of proximal legs &amp; impairment of proprioceptive sensation of legs</td>
<td>T6–8 laminectomy (NA)</td>
<td>complete recovery (4 mos)</td>
<td>NA</td>
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<td>Rahman et al., 1997</td>
<td>30, M</td>
<td>working as goldsmith</td>
<td>neck pain radiating to back of head</td>
<td>C3–4</td>
<td>pst</td>
<td>sensory disturbance below C3–4 &amp; tetraplegia</td>
<td>upper cervical laminectomy (NA)</td>
<td>complete recovery (6 mos)</td>
<td>NA</td>
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<td>Chen et al., 1997</td>
<td>50, M</td>
<td>weight lifting</td>
<td>chest tightness, upper back pain</td>
<td>T2–3</td>
<td>ant</td>
<td>paraparesis &amp; sensory disturbance below T-4</td>
<td>T2–3 laminectomy (NA)</td>
<td>partial recovery, paraparesis but muscle strength improved (8 mos)</td>
<td>NA</td>
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<td>Aitken et al., 1997</td>
<td>84, F</td>
<td>gardening</td>
<td>back &amp; chest pain radiating to shoulders</td>
<td>C4/5–T2</td>
<td>rt posterolat</td>
<td>tetraparesis</td>
<td>C4–T3 laminectomy (NA)</td>
<td>partial recovery, mild C6–7 weakness (39 days)</td>
<td>NA</td>
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<tr>
<td>Inamasu et al., 2000</td>
<td>50, M</td>
<td>working in office (businessman)</td>
<td>neck pain, rt arm numbness</td>
<td>C3–6</td>
<td>rt posterolat</td>
<td>rt sensory disturbance below C-5 &amp; rt hemiparesis</td>
<td>conservative</td>
<td>partial recovery, rt hemiparesis but muscle strength improved (4 days)</td>
<td>NA</td>
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<td>Hentschel et al., 2001</td>
<td>68, M</td>
<td>manual labor: “strenuous work on boat”</td>
<td>interscapular pain</td>
<td>C7–T1</td>
<td>pst</td>
<td>monoparesis (lt leg)</td>
<td>conservative</td>
<td>complete recovery (4 mos)</td>
<td>hematoma resolved (4 mos)</td>
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<tr>
<td>Seet et al., 2005</td>
<td>51, F</td>
<td>awakening</td>
<td>rt leg weakness, lt leg numbness</td>
<td>T10–12</td>
<td>rt anterolat</td>
<td>sensory disturbance below L1 &amp; flaccid paraplegia</td>
<td>T10–12 laminectomy (8 hrs)</td>
<td>no recovery (3 days)</td>
<td>hematoma resolved (2 wks)</td>
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<tr>
<td>Spengos et al., 2005</td>
<td>66, F</td>
<td>resting</td>
<td>lower neck pain radiating to lt arm</td>
<td>C4–7</td>
<td>lt posterolat</td>
<td>rt sensory disturbance (face spared) &amp; lt hemiparesis</td>
<td>C4–6 laminectomy (NA)</td>
<td>partial recovery, “discharged walking” (6 days)</td>
<td>NA</td>
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<td>Hsieh et al., 2006</td>
<td>20, M</td>
<td>using a computer</td>
<td>bilat shoulder pain</td>
<td>C5–7</td>
<td>rt posterolat</td>
<td>paraplegia</td>
<td>laminectomy (NA)</td>
<td>complete recovery (3 mos)</td>
<td>NA</td>
<td></td>
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<tr>
<td>Thiele et al., 2008</td>
<td>55, F</td>
<td>at work</td>
<td>weakness: “dropped her pen”</td>
<td>C4–7</td>
<td>rt posterolat</td>
<td>rt sensory disturbance (arm &amp; leg) &amp; triparesis (arms &amp; lt leg)</td>
<td>C5–6 hemilaminectomy (NA)</td>
<td>complete recovery (2 wks)</td>
<td>NA</td>
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* ant = anterior; FU = follow-up; NA = not available; pst = posterior.
The patient in our case suffered from severe occipital and nuchal pain of sudden onset followed by progressive motor paresis of the bilateral lower extremities and hands. The superficial sensations of pain and temperature were disturbed, whereas the deep sensations of vibration, position, and passive movement and the fine touch sense were completely spared. These neurological signs indicated bilateral damage to the anterior and lateral corticospinal tracts and the anterior and lateral spinothalamic tracts, but in the absence of deficits of the fasciculi gracilis and cuneatus; therefore, the clinical diagnosis in our case was anterior spinal cord syndrome. The acute onset of the neurological deficits with pain implied stroke or other vascular origin. Hence, the differential diagnosis on admission included acute ischemia or hemorrhage of the anterior spinal artery region with or without an arterial dissection. However, the MR imaging study obtained on admission showed the acute spinal epidural hematoma to be dorsal to the spinal cord.

The mechanism of the anterior spinal cord syndrome in the current case remains unclear. We suggest that the preexisting asymptomatic central-type CIDH contributed to the false localizing sign for the SSEH, namely the discrepancy between the SSEH location and the neurologically symptomatic region. We propose the following as a possible mechanism for the anterior cord syndrome in this case. First, the posterior cervical SSEH moved the spinal cord anteriorly without damaging the posterior spinal tracts. Consequently, the counterforce by the CIDH directly injured the anterior spinal cord. In addition, we could not exclude the possibility that the pinching effect due to the SSEH and CIDH modified the anterior spinal arterial blood flow. These suppositions, supported by the MR imaging findings, could well explain the neurological symptoms at the onset in our case.

No reports are available on the anterior spinal cord syndrome triggered by SSEH. Authors of an early article reported a case of cervical spinal epidural hematoma due to spinal arteriovenous malformation rupture, which was described as an anterior cord syndrome; however, deep sensation disturbance was also present. Considering these two items of information, our case is the first reported instance of posterior cervicothoracic SSEH leading to transient acute anterior spinal cord syndrome. The asymptomatic CIDH of central type was considered critical in the development of the false localizing sign for the SSEH.

Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

References


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