Spontaneous spinal epidural hematoma in a 22-month-old girl

Case report

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The authors report the case of a 22-month-old girl who developed cervical pain, neck stiffness, and quadripareisis over 12 days. An epidural hematoma was removed, with complete recovery after 6 months. There was no history of trauma. A search of the literature revealed eight previous cases of spontaneous spinal epidural hematomas in children under the age of 10 years.

Key Words • epidural hematoma • spinal hematoma • spinal cord compression • pediatric surgery

Spontaneous spinal epidural hematomas are unusual. In 1967, Markham, et al., collected 46 cases from the literature. In a review of 124 cases of spinal epidural hematomas, Piotrowski, et al., found a total of 47 cases in which no etiological factor could be identified.

The term "spontaneous" has been extended to include those hematomas that occur after apparently minimal efforts, such as lifting heavy weights, sawing wood, or pulling a chair, and in some cases the onset of symptoms occurred during sleep. Other factors reported to cause spontaneous spinal epidural hematomas include pregnancy, ankylosing spondylarthritis, arterial hypertension, and atherosclerosis. Anticoagulant therapy caused 27 of the 124 spinal epidural hematomas reviewed by Piotrowski, et al., but in these cases the term "spontaneous" is obviously inappropriate.

Spontaneous spinal epidural hematomas are extremely rare in children. We report such a case in a 22-month-old baby. Surgical evacuation resulted in complete recovery.

Case Report

This 22-month-old girl was in good health until December 9, 1980, when she started to cry continuously and became extremely irritable. On examination, the family pediatrician noticed that the child kept her head stiffly tilted to the left. Passive movement of the head was very painful. Her temperature was 37.2°C; no other signs or symptoms were noted. Grisel's syndrome (osteoarthritis of the occipitoatlantoaxial or atlantoaxial from pharyngeal infection) was diagnosed and the child was given a course of steroids. After 2 days of treatment, the child's condition had not improved, and she was admitted to the Department of Pediatrics of this hospital.

Examination. She was found to be quite alert and played as usual, although she insisted on keeping her head in the abnormal position, since the slightest movement of the neck was extremely painful. Her temperature was still normal, as were the complete blood count and sedimentation rate. Radiography of the cervical and dorsal spine showed no abnormalities.

On December 19, the child's condition deteriorated. She had trouble breathing, was unable to sit or walk, and would not move her limbs spontaneously, although she responded normally to painful stimuli. On December 21, spastic quadripareisis was noted. A lumbar puncture was made, and a complete manometric block was found to be present on jugular compression. Cerebrospinal fluid (CSF) analysis showed a protein level of 214 mg%. An emergency myelogram revealed a complete block at the C7-T1 level (Fig. 1).
Operation. A laminectomy centered on C-7 revealed a partly liquid hematoma encapsulated in a thick fibrinoid membrane, presenting the appearance of subacute subdural hematoma. The laminectomy was extended to C-5, and the collection of blood was completely evacuated. No abnormal vessels were noted. The cervical canal was noticeably dilated on the right side at the C-7 level.

Pathological study of the clot and membrane taken from the epidural space showed old blood and hemorrhagic connective tissue without abnormal vascular proliferation. Postoperative hematological studies excluded the diagnosis of a bleeding disorder. The parents could not recount any previous trauma.

Postoperative Course. The child showed progressive improvement. Three months later she could walk without assistance. Follow-up examination at 6 months showed a complete recovery.

Discussion

Spontaneous spinal epidural hematomas have been reported in only eight patients under the age of 10 years (Table 1). Five of these patients were, like our patient, babies under the age of 3 years.

The clinical signs and symptoms were always the same, that is, spinal and radicular pain at the level of the lesion, followed by more or less rapidly progressive or even acute neurological deficits. In babies, abnormal crying may initially be difficult to interpret until the development of objective neurological signs or symptoms, as was the case in our patient. The diagnosis can be achieved by the demonstration of increased CSF protein levels and/or manometric block at lumbar puncture, further confirmed by emergency myelography and operation. In the extensive review of Markham, et al., CSF abnormalities were not consistently diagnostic; they reported that the Queckenstedt test was negative in 10 of 29 cases, and CSF protein levels were normal in four of 27 cases. Therefore, the diagnosis of cord compression due to epidural hematoma should be considered principally on clinical grounds. If CSF abnormalities are lacking, a myelogram is the definitive diagnostic test.

Diagnosis is important so as to avoid the chronic course of the unfortunate child reported by Johnston. Although mild spastic sequelae have been reported in one of the nine cases summarized in Table 1, surgical decompression has provided excellent relief in all but one of these patients.

As pointed out by Ghanem and Ivan, “it is difficult to draw the line between mild trauma and the activities of daily living.” In the case reported as a spontaneous spinal epidural hematoma by Amyot, et al., there was a recent history of trauma to the shoulder. A history of trauma was also reported in Maxwell and Puletti’s chronic case in which a long period elapsed until the development of neurological symptoms, suggesting that, in young children who cannot readily express themselves verbally, some hematomas described as spontaneous might in fact be caused by a previous and maybe unknown injury. We cannot exclude this possibility in our patient. The hematoma in Jackson’s case occurred in the course of whooping cough, suggesting that a sudden increase in venous epidural pressure may have a role in the formation of this lesion. This sudden increase in pressure can also be created by such efforts as lifting heavy weights and swimming. Vascular tumors and malformations have been detected on pathological examination of...
### TABLE 1

**Summary of nine cases of spontaneous spinal epidural hematoma***

<table>
<thead>
<tr>
<th>Authors, Year</th>
<th>Age, Sex</th>
<th>Relevant History</th>
<th>Signs &amp; Symptoms</th>
<th>Manometric Block</th>
<th>CSF Protein Levels</th>
<th>Myelographic Block</th>
<th>Operation</th>
<th>Pathology</th>
<th>Recovery</th>
</tr>
</thead>
<tbody>
<tr>
<td>Johnston, 1938</td>
<td>5 yrs, M</td>
<td>skin hemangiomia below 1st scapula</td>
<td>abdominal pain, acute paraplegia, chronic paraplegia over 5 yrs</td>
<td>none</td>
<td>?</td>
<td>+</td>
<td>?</td>
<td>?</td>
<td>epidural hemangioma &amp; old fibrosis at autopsy</td>
</tr>
<tr>
<td>Shenkin, et al., 1945</td>
<td>20 mos, ?</td>
<td>&quot;acute infection-high fever&quot; 3 wks previously</td>
<td>irritability, progressive paraplegia</td>
<td>none</td>
<td>1100 mg%</td>
<td>T-5</td>
<td>+</td>
<td>?</td>
<td>?</td>
</tr>
<tr>
<td>Jackson, 1963</td>
<td>14 mos, F</td>
<td>whooping cough</td>
<td>progressive paraparesis</td>
<td>none</td>
<td>438 mg%</td>
<td>T-5</td>
<td>11th day</td>
<td>C5-T5 subacute clot</td>
<td>organizing clot</td>
</tr>
<tr>
<td>Hehman &amp; Norrell, 1968</td>
<td>21 mos, M</td>
<td>regressive irritability, staggering gait &amp; neck stiffness 6 mos previously</td>
<td>stroke his head, 12 hrs; irritability, neck stiffness, staggering gait; 5th day: spastic quadriparesis</td>
<td>+</td>
<td>170 mg%</td>
<td>T-1</td>
<td>1 mo</td>
<td>C5-T1 chronic</td>
<td>good at 15 days</td>
</tr>
<tr>
<td>Posnikoff, 1968</td>
<td>30 mos, F</td>
<td>none</td>
<td>cervicodorsal discomfort &amp; irritability; progressive quadriparesis</td>
<td>none</td>
<td>19 mg%</td>
<td>T-2</td>
<td>12 hrs</td>
<td>C7-T2 acute</td>
<td>organizing clot</td>
</tr>
<tr>
<td>Amyot, et al., 1969</td>
<td>22 mos, M</td>
<td>fell on 1st shoulder a few days previously</td>
<td>neck stiffness, 1st cervicobrachial pain, proximal 1st upper limb paresis</td>
<td>+</td>
<td>214 mg%</td>
<td>C-7</td>
<td>12th day</td>
<td>C5-T1 subacute</td>
<td>organizing clot</td>
</tr>
<tr>
<td>Ghanem &amp; Ivan, 1978 Ventureyra, et al., 1979</td>
<td>8 yrs, M</td>
<td>lifted luggage 5 days previously</td>
<td>dorsal pain after swimming; mild paraparesis improving over 3 days; 4th day: complete paraplegia</td>
<td>+</td>
<td>none</td>
<td>T-4</td>
<td>?</td>
<td>T1-3</td>
<td>organizing clot</td>
</tr>
<tr>
<td>Robertson, et al., 1979</td>
<td>6 yrs, F</td>
<td>dorsal pain of 3 wks duration</td>
<td>acute flaccid paraplegia</td>
<td>none</td>
<td>none</td>
<td>T-4</td>
<td>?</td>
<td>T1-3</td>
<td>organizing clot</td>
</tr>
<tr>
<td>Vallée, et al., 1982</td>
<td>22 mos, F</td>
<td>none</td>
<td>cervical pain, neck stiffness; progressive quadriparesis</td>
<td>+</td>
<td>214 mg%</td>
<td>C-7</td>
<td>12th day</td>
<td>C5-T1 subacute</td>
<td>organizing clot</td>
</tr>
</tbody>
</table>

* CSF = cerebrospinal fluid; + = present, ? = equivocal.

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the clot in some adult patients,6,7,12,18,26 and it is the opinion of several authors13,14,26 that vascular anomalies would be recognized more often if careful microscopic examination were employed in every case. Pathological examination was performed in six of the nine cases in Table 1; an epidural hemangioma was found at autopsy in Johnston’s case.11

### References


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