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CASE REPORT

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Spontaneous spinal epidural hematoma: an uncommon presentation of a rare disease

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Abstract *Introduction:* Spontaneous spinal epidural hematoma is rare in children. The presenting symptoms are usually pain, either local or radicular, followed by progressive bilateral weakness, and sensory loss hours and even days later. In the absence of significant precipitating factors such as severe trauma or previously known coagulopathies the diagnosis is usually delayed, and it is not until the full picture of severe cord compression is developed, that MRI is done and the diagnosis is finally made. *Case report:* We describe a case of 10-year-old girl who presented with pain and pure brachial plexus radiculopathy as the only clinical manifestations of spinal epidural hematoma. *Conclusion:* A high index of suspicion can lead to the correct diagnosis even before the development of full cord compression and thus improve the overall prognosis.

Keywords Epidural · Spinal · Spontaneous

Introduction

Spontaneous spinal epidural hematoma is an uncommon cause of cord compression in children. Although such precipitating factors as anticoagulant therapy, hemophilia, and arteriovenous malformations are common [15], 40–50% of reported cases have no identifiable cause [12]. Posttraumatic cases are relatively rare [1]. The initial presentation is usually nonspecific. The most common symptom is pain, followed by progressive symptoms of cord compression within hours or even days. We describe a case of a 10-year-old girl with cervical epidural hematoma, who presented only with pure brachial radiculopathy, with no signs or symptoms of cord compression. MRI of the spine demonstrated an epidural hematoma on the left side of the spinal canal extending from C-6 toT-1 and stretching the C-7, C-8, and T-1 nerve roots. A high index of suspicion can lead to an early surgical intervention and a better prognosis.

Case report

A 10-year-old previously healthy girl presented with a history of pain in the left arm and shoulder and weakness of her left hand. Ten days prior to admission the patient had fallen from a low bed to a carpeted floor during her night sleep, but she denied striking her head or neck. The symptoms developed the morning after, starting with shoulder pain, which was attributed to the fall, and progressing to weakness of the hand the day after, which was attributed to the pain. There was no history of a bleeding diathesis or anticoagulation treatment.

On admission the patient was afebrile, awake, and alert. Neurological examination revealed significant pain in the left arm and shoulder, which was worsened by movement, and weakness of left wrist flexion and of the intrinsic muscles of the left hand. There was decreased sensation on the inner aspect of the left hand. Deep tendon reflexes were normal. The following laboratory studies were normal: hemogram, serum electrolytes, liver function tests, coagulation studies including prothrombin time, partial thromboplastin time, bleeding time, and serum fibrinogen. MRI of the spine demonstrated an epidural hematoma on the left side of the spinal canal extending from C6 toT1 and stretching the C-7, C-8 and T-1 nerve roots and causing a mass effect with shifting of the spinal cord (Fig. 1).

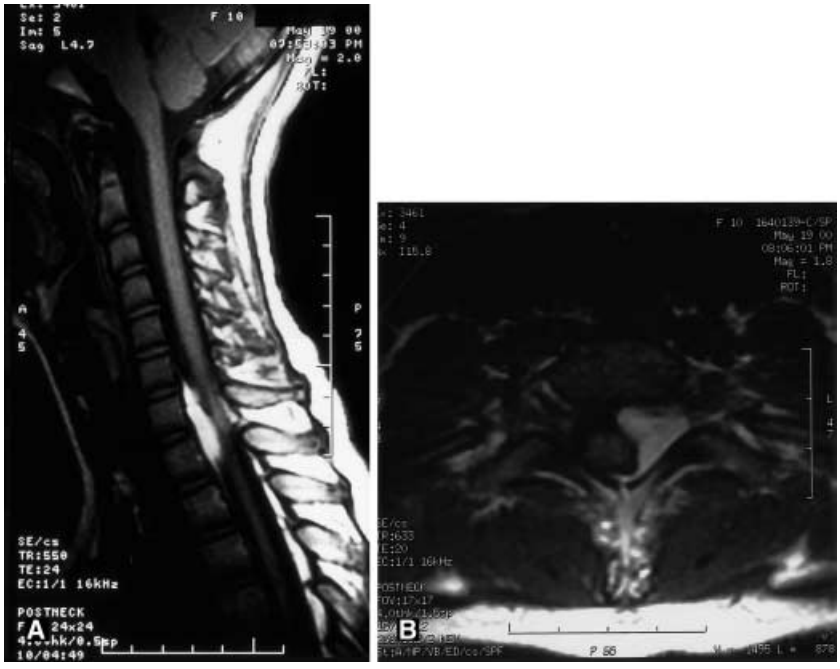

Fig. 1A, B MRI of the spine showing an epidural hematoma extending from C-6 to T-1, stretching the C-7, C-8 and T-1 nerve roots and causing a mass effect and shifting of the spinal cord. **A** Sagittal T2-weighted image (TR 550 ms, TE 24 ms). **B** Axial T2-weighted image

The patient underwent an emergent laminectomy with evacuation of the hematoma. There was no evidence of tumor or abnormal blood vessels on histological examination. The postoperative course was uneventful. The pain abated within the first 72 h, and the patient gradually regained her strength, making a full recovery during the 30 days of follow-up.

Discussion

Spontaneous spinal epidural hematoma (SSEH) is an idiopathic accumulation of blood in the vertebral epidural space; it has been reported in all age groups, but it is most frequent after the fourth or fifth decade. It is much less common in children. The clinical symptoms depend on the level of the hematoma. The most common site in children, as recently documented in 27 cases, is C-5 to T-1 [10]. This was also the level involved in our patient. The first symptom is usually pain, and it can either be localized to the level of the hematoma or be radicular in nature. In younger children the initial symptoms can be nonspecific and manifest as irritability and crying [9]. Neurological deficits such as severe weakness and a change in sensory level may develop some hours or even days after the initial pain. In most patients, MRI is done and the final diagnosis is usually made only after signs and symptoms of cord compression have already developed. The clinical presentation in our patient was different. Although pain was the initial symptom, true myelopathy never developed, and the presenting symptoms such as weakness of left wrist flexion and of the intrinsic muscles of the left-hand were more typical of brachial plexus radiculopathy.

Predisposing factors for spinal epidural hematoma are found only in 50–60% of cases. The most common include use of anticoagulants [15], coagulopathies, either congenital, such as hemophilia, or acquired, such as leukemia [8], and procedures such as spinal tap or epidural anesthesia [7]. Trauma is surprisingly rare in children, and few cases have been described [1]. It is not clear why despite the many opportunities for this rich epidural venous plexus to rupture, it so rarely does so. Cases without known predisposing factors are referred to as spontaneous. Cases in which minor trauma has occurred, but this has not been sufficiently significant to cause a bleed, are also described as spontaneous [9, 14]. In our patient the symptoms developed after the child had fallen from a low bed onto a carpeted floor at night. In the absence of any history or evidence of significant head or neck trauma, and since falls of this kind are very common in the pediatric population and are rarely associated with clinical symptoms, we suggested a diagnosis of SSEH.

Although the pathogenesis of SSEH is unclear, the bleeding is mostly believed to be venous in origin. The lack of valves in the epidural venous plexus makes it especially vulnerable to any intrinsic change in pressure. Activities such as whooping cough [6], voiding [3], weight lifting [13], and even trumpet playing [4], which can cause prolonged valsalva and sudden fluctuation in intra-abdominal and intrathoracic pressure, have been described preceding the neurological symptoms.

Other theories, such as the idea that epidural arterial hemorrhage causes mechanical disruption and traction on nerve roots or spontaneous rupture of occult arterio-

venous malformation [2] have also been suggested. No such malformations were demonstrated on our patient's MRI.

Surgical decompression and evacuation of the epidural hematoma is the treatment of choice. Complete neurological recovery was described in approximately 50% of patients and partial recovery in 44% [11]. Level of preoperative neurological deficit, severity of the neurological deficit, and operative interval have been described as the most critical factors affecting recovery [5]. Recovery was significantly better when decompression was performed less than 36 h after the onset of the neurological deficit. Children may have a better potential than adults for neurological recovery. In our patient full recovery was achieved within 30 days of surgery and was most probably secondary to the relatively mild neurological deficit and the absence of clinical signs of cord compression. A high index of suspicion can lead to an early diagnosis and intervention, and to an improvement of the overall prognosis.

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