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Spontaneous Spinal Epidural Hematoma in an Infant: A Case Report

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Abstract **Case Report**

A spontaneous spinal epidural hematoma (SSEH) is an uncommon but severe condition with a high morbidity rate, especially in infants. It results from an accumulation of blood in the spinal epidural space. It requires early diagnosis and urgent management to achieve recovery of neurologic function. It refers to a non-traumatic etiology and excludes other possible causes such as hemophilia, neoplasms, arteriovenous malformation, coagulopathies, and iatrogenic causes. The clinical features are often nonspecific, leading to a delay in diagnosis. MRI provides the most valuable visualization of the location and hematoma mass as well as the presence of the spinal cord compression. SSEH can occur in any spinal cord segment but predominantly at the posterior cervicothoracic and thoracolumbar levels. The source of hemorrhage SSEH can be both vertebral venous plexus system and arterial source. We report a rare case of an infant spontaneous spinal epidural hematoma whose diagnosis was delayed because of a recent history of angina before his development of neurologic deficits. Magnetic resonance imaging of the spine revealed a dorsal spinal epidural hematoma with compression extending from C7 to T3 a. There were no predisposing factors. Laminectomy of C7 and the first three dorsal vertebrae and complete evacuation of hematoma was done.

Keywords: Spontaneous spinal epidural hematoma- Spinal compression- Infants - Magnetic resonance imaging.

INTRODUCTION

Spontaneous spinal epidural (SSEH) is a very rare entity, especially within the pediatric population [1, 2]. It results from an accumulation of blood in the epidural space of the spinal canal that occurs in the absence of trauma, hemophilia, neoplasms, arteriovenous malformation, coagulopathies, and iatrogenic causes [3]. These hematomas can compress the spinal cord, causing acute ischemia and permanent neurological deficits [4]. Diagnosis is frequently delayed in infants due to non-specific symptomatology and limited clinical examination [5]. We are reporting one such rare case in a child.

CASE REPORT

We report the case of a 14-year-old male infant admitted to our hospital with a 5-day history of angina before his development of neck pain and progressive leg weakness resulting in an inability to stand or walk. He denied any history of trauma, systemic disease, anticoagulant usage, or coagulopathy. Neurological examination disclosed paraplegia. There was no urinary incontinence, the cranial nerves and cerebellar functions were normal. Computed tomography (CT) of the head was unremarkable. Due to concern for Guillain–Barre Syndrome (GBS), a lumbar puncture (LP) was performed which demonstrated normal inflammatory markers. Systemic examinations including hemogram, prothrombin time, and platelet count were also normal. Magnetic resonance imaging (MRI) of the spine was urgently recommended which revealed a dorsal epidural hematoma extending from C7 to T3 vertebra which appears hyperintense on T1 and T2, causing compression and anterior displacement of the spinal cord from T1 to T3 (Figure 1).

The patient underwent an emergent laminectomy extending from C7 to T3 with the evacuation of the hematoma.

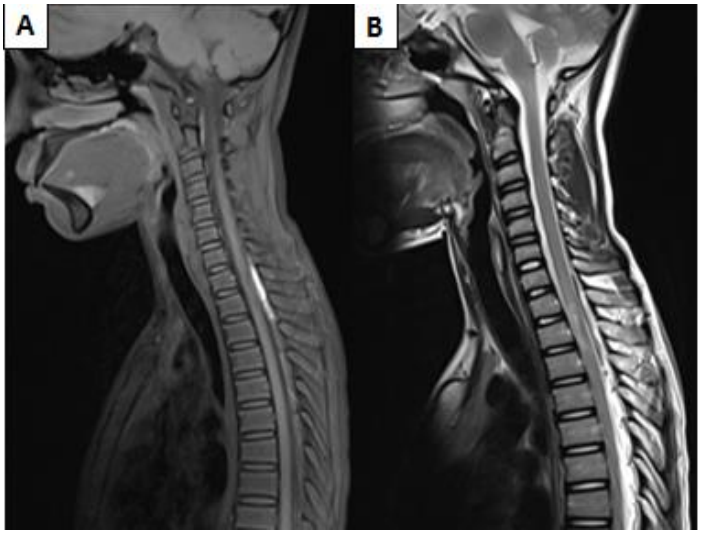

Figure 1: Sagittal T1 (A) and T2 (B) weighted MRI of the cervical spine showing a dorsal epidural hematoma extending from C7 to T3 vertebra which appears hyperintense on T1 and T2, causing compression and anterior displacement of the spinal cord from T1 to T3

DISCUSSION

Intraspinal hemorrhage first was described by DuVerney in 1688, although a later description of SSEH in a 14-year-old female by Jackson in 1869 often is considered the initial report of this entity [6, 7]. The incidence of SSEH has been reported as 0.1 per 100,000 patients in the general population and accounts for 40-50% of all reported cases of spinal epidural hematoma, but it is felt to be even less common in children and infants [3, 8]. 25-30% are associated with the use of anticoagulants, coagulopathies resulting from blood dyscrasias, such as leukemia and hemophilia, arteriovenous malformations, epidural hemangiomas, infection, trauma, and iatrogenic causes including lumbar puncture and epidural anesthesia [9].

The clinical presentation depends on the speed of accumulation of hematoma, location, and volume as estimated by the extent of spinal segments involved. Clinical characteristics of spontaneous spinal epidural hematoma in children differ significantly from those in adults [10]. Its rarity and nonspecific symptoms frequently delay diagnosis. Adults often present with sudden severe neck or back pain with associated radiculopathy and eventually progressive motor and sensory deficits. However, infants present typically in a nonspecific manner with irritability and pain with neck movement, which can precede neurologic decline for several hours or days [3, 9].

Most of the hematomas are located in the thoracolumbar region in adults, whereas involvement of the cervicothoracic region, usually C5–T1, is more common in children [10].

Bleeding in spontaneous spinal epidural hematoma is thought to result from rupture of the valveless epidural venous system [11]. The internal vertebral venous plexus is divided into anterior and posterior parts [12]. The anterior part is closely attached to the posterior longitudinal ligament by firm connective tissue strands called Hoffman ligaments and is stable [12, 13]. The posterior part courses loosely through the epidural fat, which extends laterally to surround the nerve roots. The sudden elevation of intrathoracic or intra-abdominal pressure induced by activities like crying, coughing, straining, or trauma can cause a rapid increase in backflow into this valveless venous system, with the posterior unsupported part being prone to rupture [10]. Hence, most spontaneous spinal epidural hematomas are located in the posterior aspect of the spinal canal and may extend into lateral gutters [10]. Hematoma in our patient was posterior to the cord.

Upon suspicion of SSEH, the imaging modality of choice to confirm is MRI. MRI provides the most valuable visualization of the location and hematoma mass as well as the presence of spinal cord compression [14]. Early MRI recognition provides the opportunity for early appropriate treatment and therefore leads to better neurological recovery. In the first 24 hours, hematoma usually appears isointense to the cord on T1WI and hyperintense and heterogenous on T2WI. Then hematoma usually becomes hyperintense on T1WI and T2WI during the next 48 hours. Chronic hematomas appear hypointense on both T1WI and T2WI [15]. Fat suppression images may be used to distinguish hematoma from epidural fat. Sometimes active bleeding into the hematoma will reveal a central area of enhancement when contrast is used [16].

The differential diagnosis of spontaneous spinal epidural hematoma includes an acute herniated intervertebral disc, acute ischemia of the spinal cord, epidural tumor or abscess, spondylitis, transverse

myelitis, or even a dissecting aortic aneurysm and acute myocardial infarction [17].

A decompressive laminectomy and hematoma evacuation are the standard surgical procedures upon diagnosis of SSEH. Urgent decompression of the spinal cord especially in the high-risk SSEH patients with severe neurological deficits should be the initial step for the neurological deficit recovery and the hematoma classification [18]. However, there have been several reports of SSEH treated conservatively without surgery showing spontaneous resorption or spontaneous recovery with good outcomes, especially in patients with a bleeding diathesis [3].

CONCLUSION

Spontaneous spinal epidural hematoma is extremely rare in children with nonspecific clinical features in infants. MRI is the modality of choice for diagnosis. Because SSEH is a treatable cause of neurologic decline with a prognosis that is inversely proportional to the operative interval, this diagnosis should be considered in infants with irritability and neurologic deficits, so treatment can be initiated expeditiously.

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