Case Reports

DOI: https://doi.org/10.26676/jevtm.54023

Spontaneous Spinal Epidural Hematoma in Children: Two Case Reports and a Literature Review

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Background: Spontaneous Spinal Epidural Hematoma (SSEH) is a rare emergency in the general population. It is an even rarer entity in the pediatric population.

Case Description: We report two cases: a 15-month-old boy with SSEH, who was presenting with irritability, acute pain, and a 3-year-old girl who presented with left-sided Horner's syndrome and pain in the left upper extremity. Both patients were misdiagnosed upon admission. At 10 months and 2 months, respectively, after surgery, decompressive surgery was recommended as soon as possible, but this is normally carried out 12 to 24 hours after onset.

Conclusion: The clinical presentation of SSEH is atypical and highly difficult to diagnose in children, especially in infants and toddlers. SSEH should always be included in the differential diagnosis when children present with acute back and neck pain, irritability, and uncontrolled crying.

Keywords: *Spontaneous Spinal Epidural Hematoma; Pediatric Population; Brain MRI; Case Report*

Received: 27 April 2025; Accepted: 19 May 2025

INTRODUCTION

Spontaneous spinal epidural hematoma (SSEH), also known as idiopathic spinal epidural hematoma, refers to a spontaneous or cryptogenic spinal cord compression caused by the collection of blood in the spinal epidural space without obvious predisposing factors.

Anticoagulant therapy, therapeutic thrombolysis, hemophilia and factor XI deficiency, long-time aspirin use as a platelet aggregation inhibitor, cocaine abuse, vascular malformations, and Paget disease are all suggested predisposing factors connected with SSEH [1–3].

SSEH is a rare neurosurgical emergency occurring in all age groups, especially among middle-aged and elderly patients [4]. Its age distribution shows bimodal peaks at 15 to 20 and 65 to 70 years [5]. It represents 40% to 50% of all spinal epidural hematomas with a male-to-female ratio of 1.4:1.1 [4]. The annual incidence of SSEH is estimated at an approximate 0.1 per 100,000 patients [6,7].

SSEH is mostly acute but may also be chronic, spanning months or years. According to the literature, chronic forms located in the lumbar spine are rarely described. Chronic lesions in other levels of the spine have shorter progression of neurological deficits [8].

SSEH is an even rarer entity in infants, with mostly case studies published in the literature. Within the pediatric population, especially in infants, presentation is commonly atypical and without remark. This review seeks to help raise a high index of suspicion and alertness on the side of parents, emergency physicians, and neurosurgeons to this treacherous and morbid pathology for effective and timely management.

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Ethical Approval and Informed Consent

Prior to the commencement of the study, ethical approval was obtained from the following ethical review board: The Ethics Committee of the Morozoskaya Children's City Clinical Hospital, Moscow, Russia. No. Reference L035'00115-77/00096790, 103, February 2, 2015. The CARE checklist has been followed. The study was carried out according to the latest revision of the Helsinki Declaration regarding medical research involving human subjects. The written informed consent was obtained from their parents or legal guardian. All procedures were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

CASE REPORT 1

Here, we report the case of a 15-month-old toddler who presented to our institute with acute pain, weakness, and refusal to stand or walk. His medical history was without remark. A computed tomography (CT) scan of his head revealed no intracranial pathologies. He was examined by a neurologist in the emergency department. It was not possible to examine his gait due to the child's negative response; he could not sit nor stand up on his own. Nuchal rigidity was presumed upon assessment. Muscle tone and reflexes on the right side were increased. The primary diagnosis on transfer to an infectious disease hospital was unspecified meningitis or acute post-infectious cerebellitis.

He was transferred back to our institute after he was evaluated by a magnetic resonance imaging (MRI) scan of the spine that revealed a lesion at C6–Th3. The lesion formation (suggestive of an epidural hemorrhage due to rupture of a vascular malformation) with dimensions up to 10 × 14–19 × 48 mm^2 (about 4 ml) was determined dorsally. It spread to the intervertebral foramina on the right at the C7–Th3 level and on the left at the Th2–Th3 level, without reliable signs of any solid composition.

The spinal cord was compressed and displaced anteriorly at the same level (Figure 1*a*). The spinous processes of C6–Th4 were dissected with a Misonix BoneScalpel. An epidural hematoma was observed upon dissection. Retractors were placed in between the spinous processes. The hematoma was entirely removed. The spinous processes were fixed with absorbable sutures.

Histopathological findings revealed clusters of lysed red blood cells, cellular detritus, and adipose tissue with adjacent structures resembling the walls of large-caliber vessels with pronounced deformed and fibrous walls with diffuse polymorphocellular infiltration and foci of hemorrhages. Vessels of a smaller caliber were visualized perifocally, their lumens were dilated, and the endothelium was flattened. Histochemical staining according to the Weigert method did not reveal elastic membranes in the structure of the vessel walls. These findings were consistent with spinal lymphangioma. An MRI the next day revealed complete removal of the lesion and decompression of the spine. A CT scan with contrast following the MRI showed no residual vascular malformations (Figure 1*b*). Neurological examination after surgery revealed mild left-sided hemiparesis. The patient showed a remarkable improvement in muscle strength and tone 3 months after the surgery and rehabilitation. At the 10 month follow-up, the boy could stand and walk independently with no neurological deficits.

CASE REPORT 2

A 3-year-old girl woke up in the middle of the night complaining of pain in her left hand radiating to the neck, as well as pain in the lumbar region and left half of the sternum. The next day she developed ptosis,

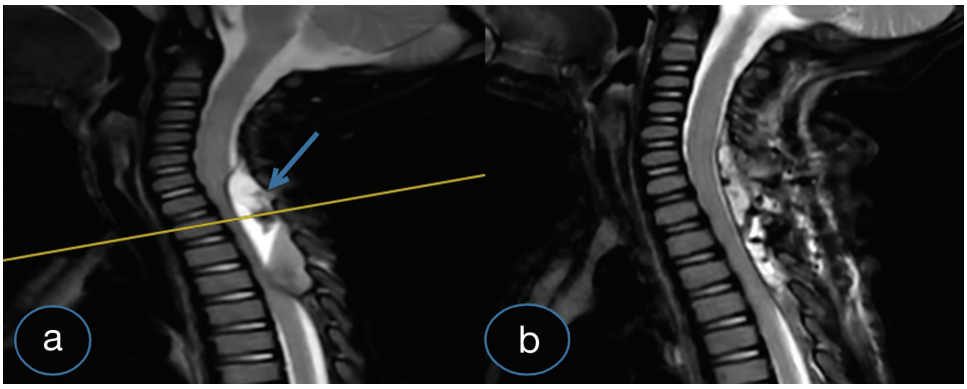

Figure 1 (**a**) Sagittal T2 weighted images using magnetic resonance imaging (MRI) revealing a dorsal epidural lesion at C6–Th3 pre-operatively. See blue arrow and yellow line. (**b**) A post-operative MRI scan showing no evidence of residual vascular malformations 10 months post-surgery.

miosis, and enophthalmos. She was examined by a pediatrician in the emergency department and was diagnosed with myalgia. She was given ibuprofen and sent back home after her parents refused admission to the hospital. On the third day, she was admitted to the ophthalmologic department after symptoms persisted and was consequently transferred to the neurological department. Her vital signs were normal and without remark. Her consciousness was clear with no cranial nerve abnormality. No nystagmus was noted. There was no papilledema on eye fundi examination. There was no deficit in motor function, muscle power, and deep tendon reflexes. The sensory examination revealed no disturbance. Laboratory tests, including a full blood cell count and coagulation profile, were without remark. Electroencephalography and electromyography findings were evaluated and found to be normal. An MRI and a contrast CT scan of the cervical spine revealed an extradural intramedullary lesion at the C7–Th4, with compression of the spinal cord (Figure 2*a*). She was urgently transferred to the neurosurgical department. The following day, the child was operated on with the help of intraoperative neurophysiological neuromonitoring. A Th1 to Th2 hemilaminectomy was performed, and 5 ml was evacuated from the epidural hematoma. A malformation of pulsating blood vessels was discovered, coagulated, and a sample was taken for histopathological examination. The pathology report showed evidence of an arteriovenous malformation. Postoperatively, deep tendon reflexes in the right upper extremity were increased. On the 2-month follow-up after surgery, the child had no neurological deficits (Figure 2*b*).

DISCUSSION

A clear pathogenesis of SSEH still remains unknown, notwithstanding the fact that there are several theories explaining the onset and course of this pathology. It is widely considered that the bleeding springs from the epidural veins. It is asserted that the valveless vertebral venous plexus burst under increased intrathoracic and intra-abdominal pressure resulting from Valsalva maneuvers, coughing, sneezing, etc. A rupture of the vessels at the so-called "locus minoris resistentiae" of the plexus in the dorsal epidural space is caused by the pressure transmitted through the abdominal and retroperitoneal veins [6,9,10]. It is worth noting that the pressure of the epidural venous plexus is lower than the intrathecal pressure. Miyagi and Kamikaseda expressed the same idea of an arterial origin of SSEH leading to the fast accumulation of blood and its accompanying neurological deficits [11,12].

In children, bleeding may be due to rupture of small vascular malformations such as venous angiomas,

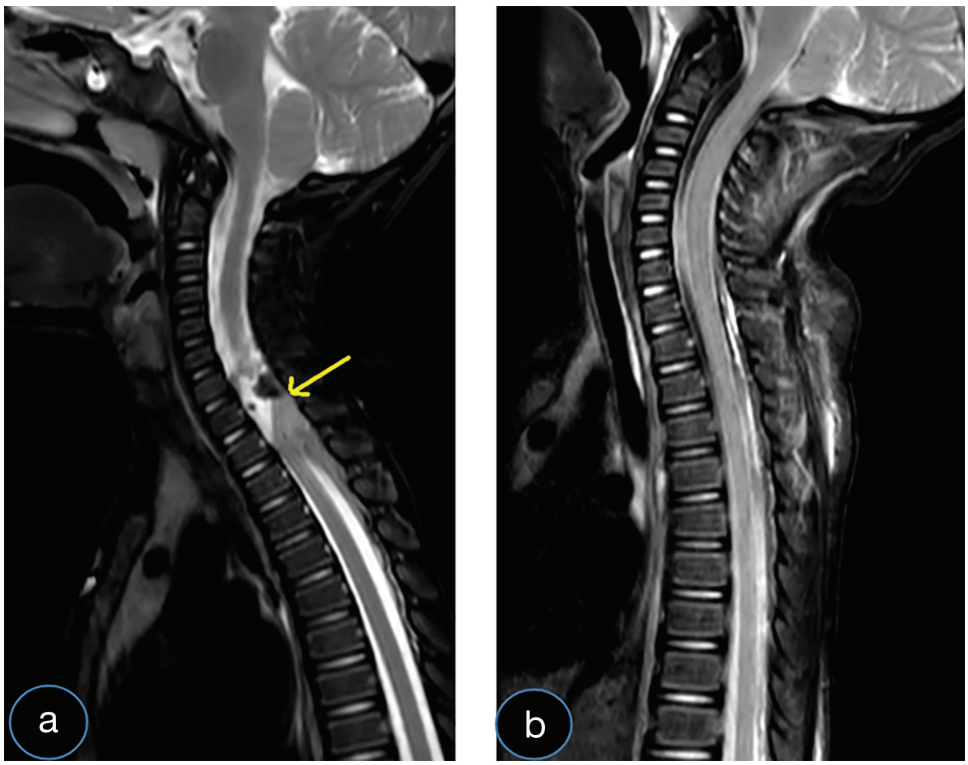

Figure 2 (**a**) Sagittal T2 weighted images using magnetic resonance imaging (MRI) showing an elongated lesion (yellow arrow) in the dorsal region of the spinal epidural space from the level of C7 to Th4 with compression of the spinal cord without edema pre-operatively. (**b**) A post-operative MRI scan with contrast showing a complete removal of the lesion 2 months post-surgery.

hemangiomas, or epidural varices. The bleeding sites in adults are C7, the lower thoracic spine, and L1. Conversely, C5 to T1 levels are the major sites of SSEH in children. The hematomas are usually located dorsally, which is anatomically congruent with the location of the venous plexus [13,14].

CLINICAL PRESENTATION AND DIAGNOSIS

Presentation of SSEH in children highly differs from the somewhat clear presentation in adults. Generally, patients may first present with acute onset of pain localized in a region of the spine and then neurological deficits such as acute or chronic spinal cord or cauda equina compression, motor and/or sensory function loss, radicular paraesthesia, and episodes of transient lower extremity paralysis. In the pediatric population, there are no concrete symptoms that point towards SSEH. Infants mainly present with irritability, restlessness, and uncontrolled crying, while older children may present with acute back pain, weakness, and paresthesia [15]. Torticollis can also be seen in infants [16,17]. The differential diagnosis includes spinal epidural abscess, pathological vertebral fracture, vascular spinal syndromes like a dissecting aortic aneurysm, intrinsic or extrinsic cord tumor, unwitnessed or minor trauma, spinal cord ischemia, disc disease, Guillain–Barré syndrome, transverse myelitis, and congenital abnormality of the spinal cord such as a syrinx [18].

ETIOLOGY

The etiologies of SSEH include blood-clotting disorders, neoplastic tumors or leukemia, vascular malformation, or spinal procedures such as lumbar puncture. In the study by Lo, it was estimated that only 11% of all intramedullary cavernous malformations discovered were present in the pediatric population. It is also thought that cavernous vascular malformations may appear at several locations in the spine of children. Children are found to present with acute symptoms more often than adults and have more rapid decompensation from their cord injury [15,19].

MRI Investigation

Avrahami et al. in their case report revealed that myelography is unnecessary, invasive, and that a CT scan provides a less suggestive image of epidural hematomas. They concluded that spinal epidural hematoma appears isointense on T1-weighted images and heterogeneous with the spinal cord in T2-weighted images within the first hours after clinical presentation. The hematoma may later become hyperintense and homogeneous in T2-weighted images. MRI of the whole spine is a gold standard for diagnosing SSEH and is taking over the place of CT with contrast. The length and extent of the epidural hematoma can also be assessed on MRI [16,20].

LABORATORY TESTS

Laboratory tests are done to exclude predisposing pathological factors and coagulation disorders. They include a full blood count, platelet count, and coagulation panel [21].

MANAGEMENT AND PROGNOSIS

SSEH is mainly managed by timely surgical decompression and evacuation of the epidural lesion followed by rehabilitation. Conservative treatment is employed when there are minimal or no neurological deficits or when evidence of an early resolution is seen. Spontaneous recovery may also occur. There are theories of fatty areolar tissue of the epidural space, which contains a rich vascular network, allowing rapid reabsorption of blood and the draining of the hematoma through the intervertebral foramen [10,22,23].

Decompressive laminectomy, which may damage the spinal ligaments, and laminotomy are employed for surgical management. It is recorded in the literature that laminectomy, especially in the pediatric population, leads to spinal instability and deformity [7,24,25]. Laminectomy with hematoma evacuation is the most effective decompressive approach for epidural hematomas. It can be single level or more depending on the extent of the hematoma [26,27].

A proper prognosis is made by thoroughly analyzing the MRI imaging, evaluating the size and length of the lesion, and the size of the spinal cord edema. Spinal cord edema may be because of the compression of draining local venous paths and circulation and is proportionate to severe neurological deficits preoperatively and unfavorable prognosis postoperatively [28,29,32].

The interval from initial ictus to surgery, the degree of neurological deficits, and the length of the lesion are key to outcome and prognosis. Lawton et al. demonstrated in their study that it was extremely difficult to provide a time range for surgical management after the onset of SSH [28–30]. However, they recommended rapid decompressive surgery for better neurological recovery [31–33].

CONCLUSION

SSEH affects all age groups, and patients generally present with acute back pain and cord compression symptoms. In the pediatric age group, specifically in infants, presentation is quite atypical and without remark. In up to 28% of all cases, the initial diagnosis is incorrect, such as in our case. MRI is the modality of choice for diagnosis. SSEH is in the cervico-thoracic region in children. Surgery within the first 12 to 24 hours provides a better prognosis, and management in children has better outcomes. Management is mainly surgical decompression followed by complete rehabilitation. A spinal angiography after surgery is highly recommended to exclude any residual lesions.

Ethics Statement

(1) All the authors mentioned in the manuscript have agreed to authorship, read and approved the manuscript, and given consent for submission and subsequent publication of the manuscript.
(2) The authors declare that they have read and abided by the JEVTM statement of ethical standards including rules of informed consent and ethical committee approval as stated in the article.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

Funding

The authors received no financial support for the research, authorship, and/or publication of this article.

Authors Contributions

Conceptualization, GGA. Methodology, software, GGA, DAES, BC. Investigation, data, GGA. Curation, surgery, FBB, NSA, DAES. Supervision, PVL, GC, EC. Visualization, validation, GM. Writing – original draft, GGA. Review and editing, formal analysis, DAES, GGA, MM, BC.

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