ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

www.surgicalneurologyint.com

Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Professor of Clinical Neurosurgery, School of Medicine, State U. of NY at Stony Brook.

SNI: Pediatric Neurosurgery

Editor
Frank Van Calenbergh, MD
University Hospitals; Leuven, Belgium

SNI
Open Access

Case Report

Spontaneous spinal epidural hematoma in infants: A case report and review of the literature

Marouane Hammoud, Hmamouche Oualid Mohammed, Khalid Chakour, Mohammed El Faiz Chaoui

Department of Neurosurgery, Medical School, University Sidi Mohammed Ben Abdellah, Hassan II University Hospital of Fez, Fez, Morocco.

E-mail: *Marouane Hammoud - marouane.hammoud@gmail.com; Hmamouche Oualid Mohammed - Oualid.hmamouche@usmba.ac.ma; Khalid Chakour - chakour.khalid@gmail.com; Mohammed El Faiz Chaoui - fmchaoui@yahoo.fr

***Corresponding author:**
Marouane Hammoud, Department of Neurosurgery, Medical School, University Sidi Mohammed Ben Abdellah, Hassan II University Hospital of Fez, Fez, Morocco.

marouane.hammoud@gmail.com

Received: 13 April 2024
Accepted: 08 August 2024
Published: 06 September 2024

DOI
10.25259/SNI_290_2024

Quick Response Code:

ABSTRACT

Background: Spontaneous spinal epidural hematomas (SSEHs) are exceedingly uncommon, especially in infants, with only two reported cases. Diagnosis can be delayed due to the nonspecificity of presenting symptoms.

Case Report: We present a case of SSEH in a 10-month-old boy admitted to the pediatric emergency department with a 5-day history of progressive lower extremity motor weakness. There was no history of prior trauma. Magnetic resonance imaging of the spine revealed a posterior epidural hematoma extending from C7 to L4. After hematoma evacuation, the patient's neurological status gradually improved, and no sensorimotor deficit was present 3 weeks postoperatively.

Conclusion: Our case suggests that surgical intervention can lead to an excellent prognosis for SSEH in infants, even if the diagnosis is delayed.

Keywords: Infant, Laminotomy, Spinal epidural hematoma, Spontaneous

INTRODUCTION

Spontaneous spinal epidural hematoma (SSEH) is an uncommon cause of cord compression, with an incidence rate of 0.1 patients/100,000/year. SSEH is predominantly observed in adults, showing a bimodal distribution with peak prevalence in the 2nd and 6th decades of life.[8]

SSEH is even rarer in the pediatric population, with only two reported infant cases in the literature.[15]

The clinical presentation of SSEH in infants varies and is mostly nonspecific. Symptoms such as crying and irritability may contribute to delayed diagnosis. However, early surgical evacuation has been reported to result in more favorable outcomes compared to adults.[3,5,12]

In this report, we present a case of a 10-month-old boy with SSEH who underwent surgical evacuation within 5 days of symptom onset. The procedure resulted in excellent outcomes, emphasizing the importance of timely surgery in managing SSEH.

CASE DESCRIPTION

A 10-month-old boy presented with a 5-day history of progressive weakness in his lower limbs. His parents denied any trauma, including shaking movements. There was no personal or family

history of bleeding or anticoagulation treatment. Neurological examination revealed symmetrical flaccid weakness in both lower limbs, hypoesthesia, and increased deep tendon reflexes bilaterally. The rest of the examination was normal.

Computed tomography (CT scan) of the spine showed no spinal fracture. However, magnetic resonance imaging (MRI) revealed a hyperacute posterior epidural hematoma extending from C7 to L4, causing spinal cord compression [Figure 1]. Coagulation studies showed no abnormalities.

To prevent spinal deformity, the patient underwent laminotomy from D6 to D9 with evacuation of the epidural hematoma. Histopathological examination of the hematoma showed no evidence of tumors, abnormal blood vessels, or vascular malformations.

One month post-surgery, the patient showed improved motor weakness and tone. Follow-up MRI showed subtotal evacuation of the hematoma [Figure 2]. In the most recent examination, conducted 1 year after surgery, the patient demonstrated further improvement and was able to stand independently. A spinal CT scan showed no deformity.

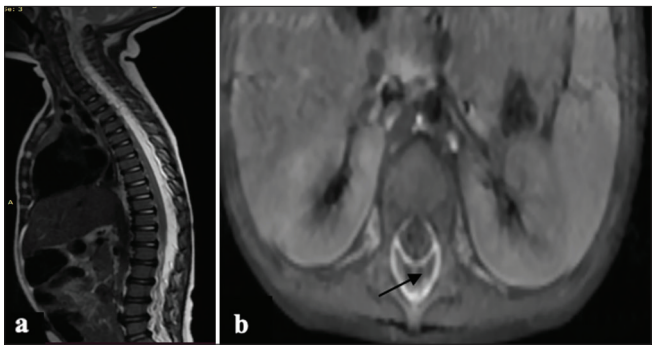

Figure 1: (a) Preoperative spinal magnetic resonance imaging (MRI). The sagittal T2-weighted image is a slightly hyperintense signal extended from C7 to L4. (b) Preoperative Spinal MRI. Axial T1-weighted image showing a posterior epidural hematoma (Black arrow).

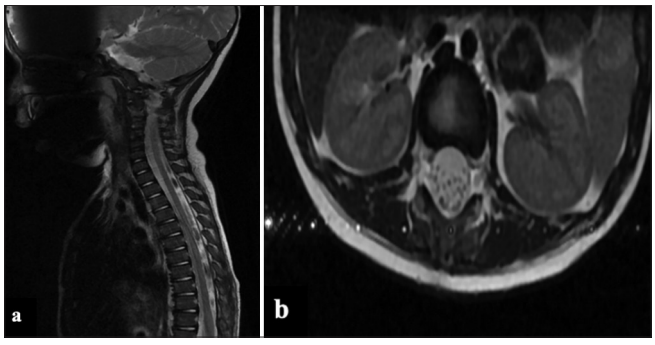

Figure 2: (a) Postoperative Spinal magnetic resonance imaging (MRI). Sagittal T2-weighted image showing subtotal hematoma resorption. (b) Postoperative spinal MRI. Axial T2-weighted image showing no compression of the dural sac.

DISCUSSION

SSEH without significant trauma in children is extremely rare. There is limited literature on this condition, mainly consisting of case reports, series, and some reviews. A systematic literature review published by Carlhan-Ledermann *et al*. in 2020 reported 153 cases of children with SSEH.[5] Among them, only three cases, including our case, have been reported in infants and ten cases in toddlers[8,12] [Table 1].

SSEH is characterized by an idiopathic accumulation of blood in the vertebral epidural space, with no known predisposing factors such as bleeding diatheses or medical procedures such as epidural and spinal injections.[1,7]

In infants, the pathogenesis of SSEH remains unclear. Most authors agree that the bleeding is venous in origin, possibly due to increased venous pressure within the intraspinal space caused by a sudden increase in intra-abdominal and thoracic pressure, leading to subsequent hemorrhage. However, in some cases, rupture of small vascular malformations such as venous angiomas, hemangiomas, or epidural varices may be the cause of bleeding.[10,16] No such malformations were detected on our patient's MRI.

The site of the hematoma in children is mainly cervicothoracic, attributed to the disproportion of head weight to body weight in the pediatric population, as well as higher cervical spine movement due to underdeveloped neck muscles.[4,10]

In our case, the site of involvement was thoracolumbar, spanning from C7 to L4.

The clinical presentation of SSEH varies depending on the speed of blood accumulation, location, and length of the hematoma. Presentation symptoms typically result from spinal cord and root compression, leading to a sudden onset of localized pain at the level of the hematoma or radicular pain. However, in infants, initial symptoms are often nonspecific, such as irritability or abnormal crying, making early diagnosis even more challenging.[11]

Symptoms generally progress rapidly; the mean duration of symptoms was 7.1 days based on a review of the previous 13 reported cases of SSEH in infants and toddlers.[2,14]

Spinal MRI is the preferred imaging modality for diagnosing and evaluating SSEH. The signal intensity on MRI varies according to the stage of the hematoma.[9]

In our case, the hematoma appeared as a low-signal intensity lesion on the T1-weighted image and as a high-signal intensity lesion on the T2-weighted image, indicating an acute hemorrhage.

Surgical evacuation as early as possible is the treatment of choice. Most authors agree that the time interval between

Table 1: Fourteen cases of infants and toddlers with spontaneous spinal hematomas in the literature.

	Sex/age (months)	Symptom to surgery (delay)	Clinical presentation	Spinal level	Outcome
Shenkin *et al*.[19]	F/20	14	Irritability, progressive Paraplegia	T1–T7	Complete recovery
Jackson[11]	F/14	NA	Progressive paraparesis	T1–T5	Complete recovery
Hehman and Norrell[10]	M/21	NA	Irritability, neck stiffness, tetraplegia	C3–T9	Marked recovery
Vallee *et al*.[20]	F/22	10	Irritability, neck pain, tetraplegia	C5–T1	Complete recovery
Licata *et al*.[14]	M/18	6	tetraparesis	T1–T2	Complete recovery
Caldarelli *et al*.[4]	F/24	3	Progressive flaccid paraplegia	C5–T4	Complete recovery
Caldarelli *et al*.[4]	F/16	9	Fever, irritability, neck pain	C4–T7	Complete recovery
Patel *et al*.[18]	M/18	7	Irritability, neck pain, tetraplegia	C1–T1	Marked recovery
Patel *et al*.[18]	F/22	NA	Fever, progressive paraparesis	C7–T10	Complete recovery
Pai and Maiya[17]	M/15	2	Weakness of upper limbs	C4–T3	Complete recovery
Liao *et al*.[15]	NA/7	NA	Irritability, crying	C2–T4	Complete recovery
Lee *et al*.[13]	M/4	5	Irritability	C4–T4	Complete recovery
Lim *et al*.[16]	F/20	14	Neck pain, paraplegia	C7–T4	Marked recovery
Our case	M/10	5	Irritability, progressive Paraplegia	C7–L4	Complete recovery

NA: Non-available, C: Cervical spine, T: Thoracic spine

onset and treatment is the primary factor influencing functional outcomes.[14] Regarding the optimal time interval for intervention, authors propose a range of 12–48 hours after symptom onset.[2,12]

However, in infants, there is often a delay in diagnosis. Surgical evacuation, even up to 3 days after onset, has been associated with good outcomes.[2,4]

Lim *et al*. reported a case in which the diagnosis and surgery were performed 2 weeks after the onset of symptoms. However, the patient demonstrated good functional recovery, suggesting that prompt surgical evacuation can still result in a satisfactory outcome for infants with delayed diagnosis.[16]

Conservative management may be considered in cases with minimal neurological deficit or cases with the early resolution of the hematoma.[9]

Regarding spinal stability outcomes, comparative studies indicate that in pediatric patients, laminotomy is preferred over laminectomy.[6,13,15]

In our case, the CT scan of the spine after 1 year showed no spinal deformity.

CONCLUSION

SSEH in infants is a rare condition with mostly nonspecific presenting symptoms, making early diagnosis challenging.

Prompt surgical decompression is crucial, irrespective of the time interval between symptom onset and operation, as it offers a better chance of recovery.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

REFERENCES

1. Abbas A, Afzal K, Mujeeb AA, Shahab T, Khalid M. Spontaneous ventral spinal epidural hematoma in an infant: An unusual presentation. Iran J Child Neurol 2013;7:47-50.
2. Alexiadou-Rudolf C, Ernestus RI, Nanassis K, Lanfermann H, Klug N. Acute nontraumatic spinal epidural hematomas. An important differential diagnosis in spinal emergencies. Spine (Phila Pa 1976) 1998;23:1810-3.
3. Al-Mutair A, Bednar DA. Spinal epidural hematoma. J Am Acad Orthop Surg 2010;18:494-502.
4. Caldarelli M, Di Rocco C, La Marca F. Spontaneous spinal epidural hematoma in toddlers: Description of two cases and review of the literature. Surg Neurol 1994;41:325-9.
5. Carlhan-Ledermann A, Laubscher B, Steinlin M, Ulrich CT,

Verma RK, Rizzi M, *et al.* Spinal epidural hematoma without significant trauma in children: Two case reports and review of the literature. BMC Pediatr 2020;20:77.
6. Domenicucci M, Mancarella C, Santoro G, Dugoni DE, Ramieri A, Arezzo MF, *et al*. Spinal epidural hematomas: Personal experience and literature review of more than 1000 cases. J Neurosurg Spine 2017;27:198-208.
7. Fok W, Sun L, Wong N, Lau P, Cheung H. Spontaneous spinal epidural haematoma in a 15-month-old boy presenting with a wry neck: A case report. J Orthop Surg (Hong Kong) 2007;15:373-5.
8. Fountas KN, Kapsalaki EZ, Robinson JS. Cervical epidural hematoma in children: A rare clinical entity. Case report and review of the literature. Neurosurg Focus 2006;20:E6.
9. Groen RJ. Non-operative treatment of spontaneous spinal epidural hematomas: A review of the literature and a comparison with operative cases. Acta Neurochir (Wien) 2004;146:103-10.
10. Hehman K, Norrell H. Massive chronic spinal epidural hematoma in a child. Am J Dis Child 1968;116:308-10.
11. Jackson FE. Spontaneous spinal epidural hematoma coincident with whooping cough. Case report. J Neurosurg 1963;20:715-7.
12. Kreppel D, Antoniadis G, Seeling W. Spinal hematoma: A literature survey with meta-analysis of 613 patients. Neurosurg Rev 2003;26:1-49.
13. Lee JS, Yu CJ, Huang KC, Lin HW, Huang CC, Chen HH. Spontaneous spinal epidural hematoma in a 4-month-old infant. Spinal Cord 2006;3:1-5.
14. Licata C, Zoppetti MC, Perini SS, Bazzan A, Gerosa M, Da Pian R. Spontaneous spinal haematomas. Acta Neurochir (Wien) 1988;95:126-30.
15. Liao CC, Lee ST, Hsu WC, Chen LR, Lui TN, Lee SC. Experience in the surgical management of spontaneous spinal epidural hematoma. J Neurosurg 2004;100:38-45.
16. Lim JJ, Yoon SH, Cho KH, Kim SH. Spontaneous spinal epidural hematoma in an infant: A case report and review of the literature. J Korean Neurosurg Soc 2008;44:84-7.
17. Pai SB, Maiya PP. Spontaneous spinal epidural hematoma in a toddler-a case report. Childs Nerv Syst 2006;22:526-9.
18. Patel H, Boaz JC, Phillips JP, Garg BP. Spontaneous spinal epidural hematoma in children. Pediatr Neurol 1998;19:302-7.
19. Shenkin HA, Horn RC, Grant FC. Lesions of the spinal extradural space producing cord compression. Arch Surg 1945;51:125-46.
20. Vallee B, Besson G, Gaudin J, Person H, Le Fur JM, Le Guyader J. Spontaneous spinal epidural hematoma in a 22-month-old girl. J Neurosurg 1982;56:135-8.

How to cite this article: Hammoud M, Mohammed HQ, Chakour K, Chaoui ME. Spontaneous spinal epidural hematoma in infants: A case report and review of the literature. Surg Neurol Int. 2024;15:320. doi: 10.25259/SNI_290_2024