Case Report

Spontaneous spinal epidural hematoma in children: a case report and literature review

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Background: Spontaneous spinal epidural hematoma (SSEH) is a hematoma within the spinal epidural space without the underlying causes of trauma or iatrogenic and is considered a very rare neurosurgical emergency disease in children that can cause spinal cord compression and neurological dysfunction. This article provides useful information and guidance to the clinician about SSEH in children regarding its specific characteristics, clinical presentation, and management strategy to achieve a better outcome.

Case Description: A 14-year-old boy presented with an acute onset of neck pain radiating to the right shoulder and progressive right hemiparesis. The cervical spine magnetic resonance imaging (MRI) revealed a right posterolateral hyperacute spinal epidural hematoma at C4–C7. The patient underwent an emergent open-door laminoplasty (C5–C6) with partial laminectomy (C4 and C7) and complete evacuation of the hematoma. The patient had a complete recovery after surgery with no neurological deficits. A literature search in the PubMed electronic database was performed to identify published English articles between January 2000 to December 2023 focusing on SSEH in children. We have found 81 articles with a total of 95 cases of SSEH in children, providing comparison data on sex, age, clinical presentation, etiology, location of the hematoma, treatment modalities, and outcomes.

Conclusions: SSEH in children is a very rare neurosurgical emergency disease. Prompt and proper examination is essential to establish the diagnosis and early surgical decompression. Adequate surgical decompression may reduce intradural pressure and increase the blood perfusion to the spinal cord, thus, this will eventually reduce ischemia and prevent secondary spinal injury. As a result, complete recovery can be expected.

Keywords: Spontaneous spinal epidural hematoma (SSEH); children; cervical laminoplasty; case report

Submitted Apr 24, 2024. Accepted for publication Jul 17, 2024. Published online Aug 26, 2024.
doi: 10.21037/jss-24-49
View this article at: https://dx.doi.org/10.21037/jss-24-49

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Introduction

Spontaneous spinal epidural hematoma (SSEH) in children is considered a very rare neurosurgical emergency case that can cause spinal cord compression and neurological dysfunction. The incidence is estimated to be 0.1 patients per 100,000 patients per year in the adult population, while the incidence in children is still unknown (1,2). The scarcity of incidence in the children population is indicated by few SSEH reports on children (3). This article focuses on SSEH in children aged 18 years old and below because of its rarity in the literature and its specific characteristics different from adults.

SSEH is defined as hematoma within the spinal epidural space without the underlying causes of trauma or iatrogenic. Most of the underlying causes of SSEH in children are idiopathic but sometimes, it can also be due to identifiable causes such as bleeding disorders, vascular malformations, and anticoagulant therapy (4-6). The spinal compression due to SSEH may potentially cause the spinal cord or nerve root injury causing permanent neurological dysfunction which demands prompt and proper management (2,7).

We present a case of a 14-year-old boy having cervical SSEH with a very rare symptom mimicking stroke and a literature review of 81 articles with a total of 95 reported cases of SSEH in children between January 2000 to December 2023. The search strategy is summarized in *Table 1*. The details of these reviewed cases are available in tabular form in the Supplementary file (Appendix 1). We provide a summary of the comparison data on sex, age, clinical presentation, etiology, location of the hematoma, treatment modalities, and outcomes (*Table 2*). We present this article in accordance with the CARE reporting checklist (available at https://jss.amegroups.com/article/view/10.21037/jss-24-49/rc).

Highlight box

Key findings

- A 14-year-old boy presented with an acute onset of neck pain radiating to the right shoulder and progressive right hemiparesis was diagnosed with spontaneous spinal epidural hematoma (SSEH) at C4–C7. The patient was treated with cervical laminoplasty and hematoma evacuation. Post-operative outcome showed a complete recovery with no neurological deficits.

What is known and what is new?

- SSEH in children is considered a very rare neurosurgical emergency case that can cause spinal cord compression and neurological dysfunction. SSEH is defined as hematoma within the spinal epidural space without the underlying causes of trauma or iatrogenic.
- We report a rare case with an unusual presentation and review of the literature of 95 reported cases of SSEH in children between January 2000 to December 2023, providing comparison data on sex, age, clinical presentation, etiology, location of the hematoma, treatment modalities, and outcomes.

What is the implication, and what should change now?

- This article provides useful information and guidance to the clinician about SSEH in children regarding its specific characteristics, clinical presentation, and management strategy to achieve a better outcome.

Case presentation

A 14-year-old previously healthy boy came to our hospital with the complaint of acute onset of posterior neck pain during singing karaoke 2 hours prior to arriving at the emergency room. There was no history of trauma on the head or spine, anticoagulant therapy, bleeding disorders, or other systemic complaints. His pain was present from the neck radiating to the right shoulder and did not improve. Before coming to the hospital, he developed a progressive weakness on the right side of his body and was not able to stand without support.

On admission, he was alert and hemodynamically stable. There was pain in the posterior neck evaluated by the visual analog scale with 7 points of 10. Neurological examination revealed right hemiparesis with manual muscle test (MMT) of 3/5 in the right extremities whereas normal on the left extremities without sensory deficits and bladder-bowel dysfunction. Laboratory blood examinations, including a thorough evaluation of the coagulation profile, were within normal range. Urgent brain and spine magnetic resonance imaging (MRI) were performed. The brain MRI result showed neither infarction, hematoma, or dissection lesions. The spine MRI revealed a right posterolateral epidural lesion in C4–C7 levels which appeared isointense on T1-weighted images (T1WI) and hyperintense on T2-weighted images (T2WI) consistent with a hyperacute spinal epidural hematoma (SEH) that caused anterior spinal cord compression to the left (*Figure 1*).

The patient underwent an emergent open-door laminoplasty (C5–C6) with partial laminectomy (C4 and C7) and evacuation of the hematoma (*Figure 2*). Intraoperative findings seemed to be no tumors or vascular abnormalities. After the hematoma was completely evacuated, the dorsal spinal cord appeared pulsatile indicating adequate

Table 1 The search strategy summary

Items	Specification
Date of search	01 January 2024
Databases and other sources searched	PubMed
Search terms used	"Spontaneous", "spinal epidural hematoma", "pediatric", "children"
Timeframe	January 2000–December 2023
Inclusion and exclusion criteria	Inclusion: case report or case series articles of SSEH in children (neonates to 18 years old), with detailed information on sex, age, clinical presentation, etiology, location of the hematoma, treatment modalities, and outcomes
	Exclusion: articles not including children or lacking sufficient detail
Selection process	The selection process was done manually by all authors

SSEH, spontaneous spinal epidural hematoma.

decompression. The follow-up cervical CT scan 7 days after surgery revealed no residual hematoma (*Figure 3*). Spinal angiography was also performed to exclude spinal vascular malformation and showed no abnormality. His symptoms were gradually improved postoperatively. On day 1 after surgery, MMT on the right extremities improved to 4/5. The patient had a complete recovery on day 5 after surgery with no neurological deficit.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Informed consent was taken from the patient's guardians for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

In this article, SSEH refers to all SEH that is not caused by trauma which is in accordance with most of the authors. Some authors only refer to SSEH if the cause is idiopathic or unidentifiable (8,9). Others also differentiate between spontaneous and idiopathic SEHs (10). SSEH in children is very rare but clinically very important due to its compressive effect on the spinal nerve and potential permanent neurological deficits. Kreppel *et al.* have reported that SEHs were the most frequent type of spinal hematoma cases (74.2%) (10).

Children with SSEH have specific characteristics that are different from adults. Younger children may have difficulty expressing their symptoms clearly compared to adults, potentially leading to delays in diagnosis (11). SSEH in children may be associated with certain underlying conditions, such as bleeding disorders or vascular malformations, which are less common in adults (6). Imaging studies such as MRI are typically used for diagnosis, but sedation or anesthesia may be required in younger children, adding complexity to the diagnostic process. Treatment of SSEH in children may require specialized care tailored to their age, size, and underlying medical conditions.

Etiology

The etiology of SSEH in children is mostly idiopathic, but there are some reports of underlying causes including hemophilia, vascular malformation, and coagulopathy. There are two well-known pathogenesis of SSEH including venous origin and arterial origin. The epidural venous plexus has a thin wall and valveless making it vulnerable to any increase of intravenous pressure. Due to its continuity with the abdominal and thoracic venous system, an increase in intraabdominal or intrathoracic pressure can also increase the intravenous pressure in the epidural venous plexus which may cause venous plexus rupture and bleeding at the posterior epidural space in most of the cases (8,12-15). Beatty (16) theorized that the free epidural artery is the source of acute SSEH in the cervical. They insisted that the arterial plexus on the epidural space is anatomically vulnerable to certain mechanical movements that may overstretch the free arteries exceeding the tolerance limitation and eventually causing arterial rupture (12,16). The possible mechanism in our case is a sudden increase of intra-abdominal or intrathoracic pressure during singing karaoke which also increases the intravenous pressure of the spinal epidural

Table 2 Patient characteristics summary

Characteristics	Total cases (%)
Sex	
Male	60 (63.2)
Female	32 (33.7)
N/A	3 (3.2)
Age (years) (average 6.7 years ±5.5 SD)	
<1	22 (23.2)
1–6	29 (30.5)
7–12	27 (28.4)
13–18	17 (17.9)
Clinical presentation	
Paraparesis	45 (47.4)
Neck pain	27 (28.4)
Tetraparesis	23 (24.2)
Back pain	21 (22.1)
Irritability	18 (18.9)
Urinary symptoms	18 (18.9)
Neck stiffness	15 (15.8)
Paresthesia	14 (14.7)
Monoparesis	7 (7.4)
Extremity pain	4 (4.2)
Horner syndrome	4 (4.2)
Hemiparesis	3 (3.2)
Ataxia	2 (2.1)
Etiology	
Idiopathic	47 (49.5)
Hemophilia	29 (30.5)
Vascular malformation	16 (16.8)
Coagulopathy	3 (3.2)
Location (average 7.5 spinal level ±6.3 SD)	
Cervical	74 (77.9)
Thoracal	82 (86.3)
Lumbar	12 (12.6)
Sacrum	6 (6.3)
Anterior/posterior location	
Posterior	74 (77.9)
Anterior	12 (12.6)
Both	2 (2.1)
N/A	7 (7.4)
Treatment procedure	
Laminectomy	37 (38.9)
Hemilaminectomy	14 (14.7)
Laminoplasty	10 (10.5)
Laminotomy	5 (5.3)
Embolization	4 (4.2)
Conservative	7 (7.4)
Factor replacement	29 (30.5)
Treatment type	
Surgery only	55 (57.9)
Surgery and factor replacement	8 (8.4)
Surgery and embolization	3 (3.2)
Non-surgical	28 (29.4)
N/A	1 (1.1)
Outcomes	
Complete recovery	65 (68.4)
Partial recovery	17 (17.9)
No recovery	10 (10.5)
Death	2 (2.1)
N/A	1 (1.1)

This table presents summarized data on SSEH in children published in the literature from January 2000 to December 2023. The data are categorized based on demographics, clinical presentation, etiology, hematoma location, treatment methods, and outcomes. The data are presented in terms of number of cases and percentage in each category. SD, standard deviation; N/A, not available; SSEH, spontaneous spinal epidural hematoma.

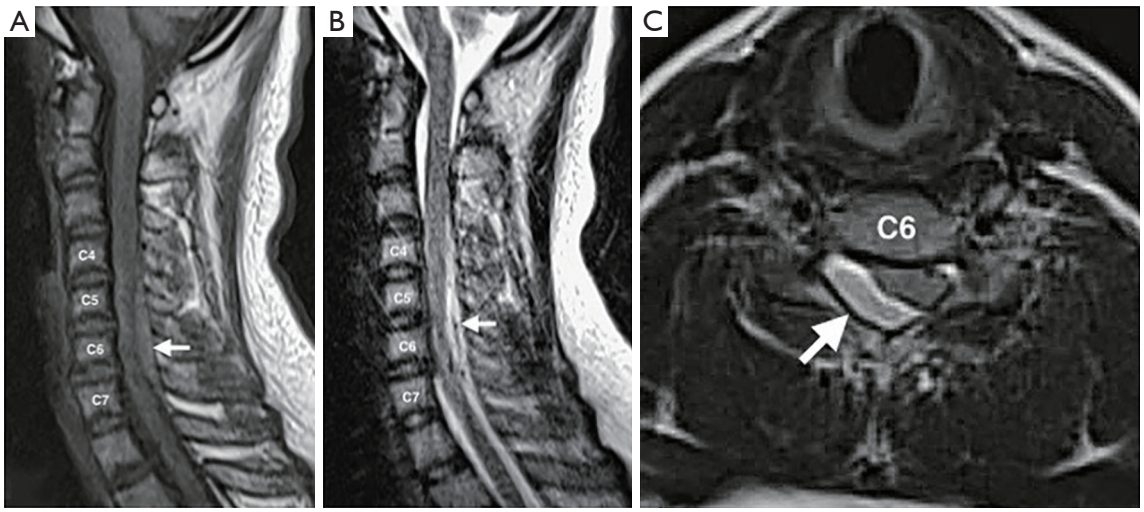

Figure 1 Cervical spine MRI showing SEH (white arrows) from C4 to C7 levels. (A) T1-weighted sagittal view; the SEH appeared isointense. (B) T2-weighted sagittal view; the SEH appeared hyperintense. (C) T2-weighted axial view at the level of C5/6 foramen; the SEH located at the right posterolateral compressing anteriorly the spinal cord to the left. MRI, magnetic resonance imaging; SEH, spinal epidural hematoma.

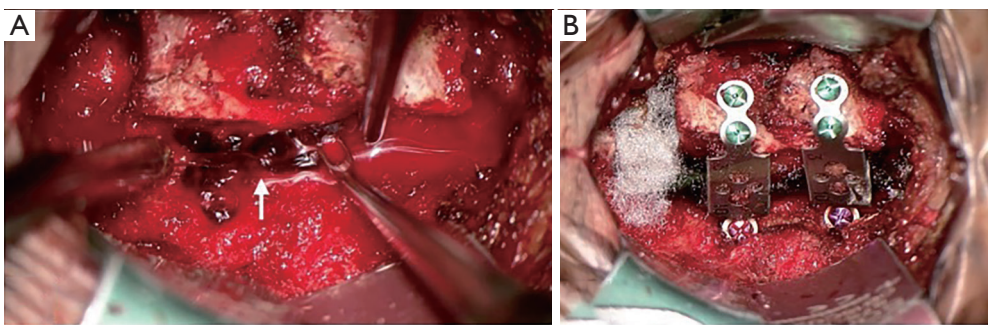

Figure 2 Open-door laminoplasty C5–C6. (A) SEH (white arrow) is exposed on the right side of the lamina. (B) Cervical laminoplasty using the laminoplasty plates. SEH, spinal epidural hematoma.

veins eventually bleeding occurs. The patient then developed a progressive hemiparesis.

Clinical presentation

Symptoms and neurological deficits of SSEH cases vary depending on several factors such as age, underlying etiology, and location of the hematoma which is the most important factor. In younger patients, the symptoms are often non-specific which makes it difficult to make the diagnosis and to identify the underlying etiology. These non-specific signs and symptoms may cause delays in the diagnosis and treatment (11). Some infant patients with SSEH had complaints of general clinical presentations such as irritability (3,17-24) and torticollis (11,18,23-25). The neurological deficit due to SSEH generally occurs within a few minutes or hours by the expanded hematoma but some can appear after a few days (26). Older children can explain their complaints which makes it easier to assess compared to infant patients.

The most common clinical presentations are weakness in the lower extremities, weakness in all four extremities, neck pain, and back pain. Some also present with uncommon clinical features such as Horner syndrome (27,28), a clinical presentation like that of a spinal birth injury (29), and hemiparesis (19,22,26) mimicking stroke like in our

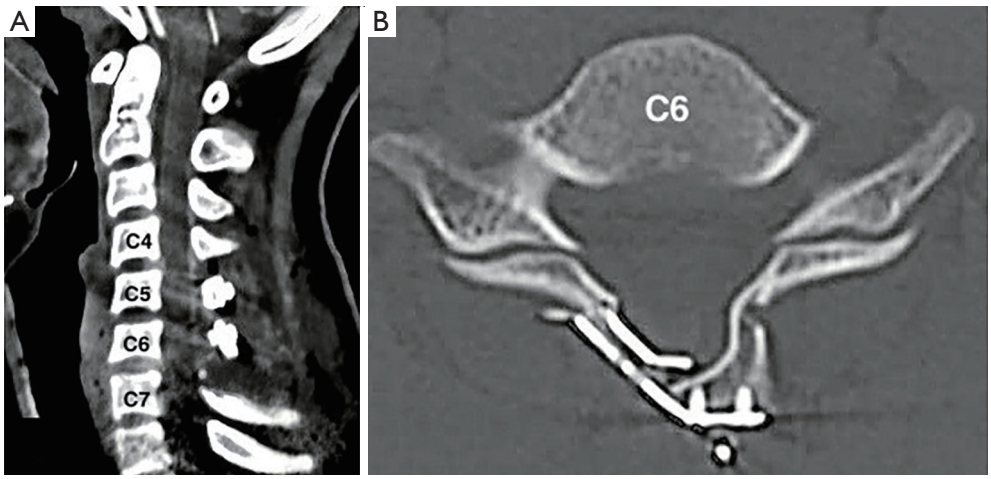

Figure 3 Post-operative CT scan. (A) Sagittal view; showed no residual hematoma in C4–C7. (B) Axial view of C6; laminoplasty using laminoplasty plates and well-decompressed spinal canal. CT, computed tomography.

present case. The clinical manifestations can also mimic other medical conditions such as meningitis (3,26,30,31), Guillain Barre syndrome (18,32,33), infections (20,27), and neoplasms.

The symptom of neck pain radiating to the right shoulder in our patient is an early clinical sign of spinal or nerve root compression. MRI should be performed promptly to avoid further myelopathy, particularly among children or young adults. SSEH should always be considered in similar situations (26). Early identification of the symptoms and work-up diagnosis can prevent delays in the management of SSEH.

Diagnosis

Although SSEH is a rare diagnosis, a high index of suspicion for SSEH should always be considered in the appropriate clinical presentation. The sudden onset of neck pain or back pain and progressive neurological deficit should make SSEH one of the differential diagnoses and is an indication for an emergent spinal MRI examination (34). Prompt identification of the symptoms and rapid diagnosis evaluation are important factors in reducing the delay to surgical decompression (35). In our patient, the course of symptoms was unusual. Because of the rapid workup diagnosis and early detection of cervical SSEH, we can perform surgical decompression without any delay which also contributes to the patient's good outcome.

MRI is considered better compared to other radiographical in revealing the location of the hematoma and the extent of spinal cord compression, as well as the consistency and stage of the hematoma (6,25,36). The hematoma in our patient appears isointense on T1WI and hyperintense on T2WI indicating a hyperacute SEH with an onset time of less than 24 hours. MRI may also show abnormally enlarged blood vessels suspected of spinal vascular malformation although MRI may not always rule out vascular malformations. Spinal angiography is an essential diagnostic tool if there is suspicion of vascular malformation or in negative cases where the MRI findings cannot reveal small vascular lesions (8,9,25,37).

Treatment

Surgical decompression is the main treatment for SSEH. The available time to restore the neurological deficit is limited, therefore early surgical decompression will give an optimal chance for good functional recovery (5,38). Laminectomy with hematoma evacuation is the most common and effective decompressive procedure in SSEH treatment, however, there are some potential problems in pediatric patients because there is a risk of progressive kyphotic deformity (6). Consequently, hemilaminectomy (6,9,13,39-49), laminotomy (29,50-53), and laminoplasty (27,33,54-59) have been used as alternative options. In our case, we performed C5–C6 open-door laminoplasty with partial laminectomy at C4 and C7. The advantage of laminoplasty is that it can preserve the major posterior structure, while still allowing for spinal canal decompression.

Conservative or non-surgical treatment of SSEH can

be an option in a mild neurological deficit or bleeding disorder. However, the option for conservative treatment of SSEH may also be considered in cases that coexist with serious coagulopathy or high risk for surgical treatment and is not always based on the mild clinical course alone (6,38,60). During the conservative treatment, there is still a possibility of neurological deterioration and enlargement of the hematoma. Therefore, close neurological monitoring and repeated MR imaging are necessary (15,61).

In SSEH due to bleeding disorder (hemophilia A or B), treatment with replacement clotting factor VIII (11,19-24,28,41,49,56,62-70) or factor IX (17,71-74) has resulted in the resolution of the hematoma without surgical intervention (61). Surgical resection and endovascular embolization have been reported as the treatment of SSEH with spinal arteriovenous malformation (4,9,33,36,43,57,75,76) and spinal dural arteriovenous fistula (30,77).

Outcome

The main contributing factors to the neurological outcome following SSEH treatment are the location of the hematoma (level of vertebral segments involved), the preoperative neurological condition, and the time interval from the onset to surgical decompression. Surgery performed in less than 36 hours in patients with a complete preoperative sensorimotor deficit and less than 48 hours in patients with an incomplete preoperative sensorimotor deficit are both associated with favorable outcomes (35). In our case, the patient had undergone surgery less than 12 hours from the onset. The post-surgical outcome showed a very good result. Early decompression is important and has shown a significant improvement statistically and clinically (78). Adequate surgical decompression may reduce intradural pressure and increase the blood perfusion to the spinal cord, this will eventually reduce ischemia and prevent secondary injury from occurring (79).

Several reports suggest that when the symptoms are mild and improve spontaneously after a few hours before surgery, a favorable outcome can be achieved without surgery (18,80). In hemophilia patients with SSEH, conservative treatment with clotting factor replacement has shown good outcomes (17,19-24,28,62,65-73), instead of performing high-risk surgical treatment with abnormal coagulation status unless neurological deterioration progresses rapidly (17).

Conclusions

SSEH in children is a very rare neurosurgical emergency that can cause spinal cord compression and neurological dysfunction. Prompt and proper examination is essential to establish the diagnosis and early surgical decompression. Adequate surgical decompression may reduce intradural pressure and increase the blood perfusion to the spinal cord, thus, this will eventually reduce ischemia and prevent secondary spinal injury. As a result, complete recovery can be expected.

Acknowledgments

Funding: None.

Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://jss.amegroups.com/article/view/10.21037/jss-24-49/rc

Peer Review File: Available at https://jss.amegroups.com/article/view/10.21037/jss-24-49/prf

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://jss.amegroups.com/article/view/10.21037/jss-24-49/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Informed consent was taken from the patient's guardians for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

References

1. Holtås S, Heiling M, Lönntoft M. Spontaneous spinal epidural hematoma: findings at MR imaging and clinical correlation. Radiology 1996;199:409-13.
2. Soltani S, Nogaro MC, Rougelot C, et al. Spontaneous spinal epidural haematomas in children. Eur Spine J 2019;28:2229-36.
3. Lee JS, Yu CY, Huang KC, et al. Spontaneous spinal epidural hematoma in a 4-month-old infant. Spinal Cord 2007;45:586-90.
4. Kirwan R, Saigal G, Faingold R, et al. Nontraumatic acute and subacute enhancing spinal epidural hematoma mimicking a tumor in a child. Pediatr Radiol 2004;34:499-502.
5. Abram HS, DeLaHunt MJ, Merinbaum DJ, et al. Recurrent spontaneous spinal epidural hematoma in a child: first case report. Pediatr Neurol 2007;36:177-80.
6. Babayev R, Ekşi MŞ. Spontaneous thoracic epidural hematoma: a case report and literature review. Childs Nerv Syst 2016;32:181-7.
7. Endo T, Suzuki S, Inoue T, et al. Prediction of neurological recovery in spontaneous spinal epidural hematoma using apparent diffusion coefficient values. Spinal Cord 2014;52:729-33.
8. Schoonjans AS, De Dooy J, Kenis S, et al. Spontaneous spinal epidural hematoma in infancy: review of the literature and the "seventh" case report. Eur J Paediatr Neurol 2013;17:537-42.
9. Carlhan-Ledermann A, Laubscher B, Steinlin M, et al. Spinal epidural hematoma without significant trauma in children: two case reports and review of the literature. BMC Pediatr 2020;20:77.
10. Kreppel D, Antoniadis G, Seeling W. Spinal hematoma: a literature survey with meta-analysis of 613 patients. Neurosurg Rev 2003;26:1-49.
11. Cuvelier GD, Davis JH, Purves EC, et al. Torticollis as a sign of cervico-thoracic epidural haematoma in an infant with severe haemophilia A. Haemophilia 2006;12:683-6.
12. Halim TA, Nigam V, Tandon V, et al. Spontaneous cervical epidural hematoma: report of a case managed conservatively. Indian J Orthop 2008;42:357-9.
13. Zhong W, Chen H, You C, et al. Spontaneous spinal epidural hematoma. J Clin Neurosci 2011;18:1490-4.
14. Abbas A, Afzal K, Mujeeb AA, et al. Spontaneous ventral spinal epidural hematoma in an infant: an unusual presentation. Iran J Child Neurol 2013;7:47-50.
15. Raasck K, Habis AA, Aoude A, et al. Spontaneous spinal epidural hematoma management: a case series and literature review. Spinal Cord Ser Cases 2017;3:16043.
16. Beatty RM, Winston KR. Spontaneous cervical epidural hematoma. A consideration of etiology. J Neurosurg 1984;61:143-8.
17. Iwamuro H, Morita A, Kawaguchi H, et al. Resolution of spinal epidural haematoma without surgery in a haemophilic infant. Acta Neurochir (Wien) 2004;146:1263-5.
18. Liao CC, Lee ST, Hsu WC, et al. Experience in the surgical management of spontaneous spinal epidural hematoma. J Neurosurg 2004;100:38-45.
19. Kubota T, Miyajima Y. Spinal extradural haematoma due to haemophilia A. Arch Dis Child 2007;92:498.
20. Kalina P, Morris J, Raffel C. Spinal epidural hematoma in an infant as the initial presentation of severe hemophilia. Emerg Radiol 2008;15:281-4.
21. Kiehna EN, Waldron PE, Jane JA. Conservative management of an acute spontaneous holocord epidural hemorrhage in a hemophiliac infant. J Neurosurg Pediatr 2010;6:43-8.
22. Tagaya A, Miyajima Y, Sakamoto M, et al. Spontaneous spinal epidural hematoma in an infant with undiagnosed hemophilia A. Pediatr Int 2010;52:296-8.
23. Oymak Y, Muminoglu N, Ay Y, et al. Torticollis in a haemophilic infant with inhibitor: a case of spinal epidural haematoma. Blood Coagul Fibrinolysis 2016;27:576-9.
24. Jang HN, Yoon HS, Park YS. Management of spontaneous spinal epidural hematoma developed in children with hemophilia. Clinical Pediatric Hematology-Oncology 2018;25:180-4.
25. van Heesewijk JP, Casparie JW. Acute spontaneous spinal epidural haematoma in a child. Eur Radiol 2000;10:1874-6.
26. Jumani DB, Littlewood R, Iyer A, et al. Spontaneous spinal epidural haematoma mimicking meningitis in a 2-year-old child--a case report and literature review. Childs Nerv Syst 2013;29:1795-8.
27. Kwasnicki A, Calandriello A, Nikas D. Spontaneous spinal epidural hematoma in an infant presenting with Horner syndrome. Childs Nerv Syst 2022;38:827-30.
28. Morsing IE, Brons P, Th Draaisma JM, et al. Hemophilia a and spinal epidural hematoma in children. Neuropediatrics 2009;40:245-8.
29. Blount J, Doughty K, Tubbs RS, et al. In utero

spontaneous cervical thoracic epidural hematoma imitating spinal cord birth injury. Pediatr Neurosurg 2004;40:23-7.
30. Chuang NA, Shroff MM, Willinsky RA, et al. Slow-flow spinal epidural AVF with venous ectasias: two pediatric case reports. AJNR Am J Neuroradiol 2003;24:1901-5.
31. Romagna A, Potthast F, Steinborn MM, et al. Surgical treatment of a spontaneous spinal epidural hematoma under antithrombotic treatment of a multisystem inflammatory syndrome in children associated with SARS-COV-2. Childs Nerv Syst 2022;38:1213-6.
32. Lo MD. Spinal cord injury from spontaneous epidural hematoma: report of 2 cases. Pediatr Emerg Care 2010;26:445-7.
33. Paraskevopoulos D, Magras I, Polyzoidis K. Spontaneous spinal epidural hematoma secondary to extradural arteriovenous malformation in a child: a case-based update. Childs Nerv Syst 2013;29:1985-91.
34. Baeesa S, Jarzem P, Mansi M, et al. Spontaneous Spinal Epidural Hematoma: Correlation of Timing of Surgical Decompression and MRI Findings with Functional Neurological Outcome. World Neurosurg 2019;122:e241-7.
35. Groen RJ, van Alphen HA. Operative treatment of spontaneous spinal epidural hematomas: a study of the factors determining postoperative outcome. Neurosurgery 1996;39:494-508; discussion 508-9.
36. Min S, Duan Y, Jin A, et al. Chronic spontaneous cervicothoracic epidural hematoma in an 8-month-old infant. Ann Saudi Med 2011;31:301-4.
37. Moriarty HK, O Cearbhaill R, Moriarty PD, et al. MR imaging of spinal haematoma: a pictorial review. Br J Radiol 2019;92:20180532.
38. 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.
39. Rosenberg O, Itshayek E, Israel Z. Spontaneous spinal epidural hematoma in a 14-year-old girl. Case report and review of the literature. Pediatr Neurosurg 2003;38:216-8.
40. Fok WM, Sun LK, Wong NM, et al. 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.
41. Rois PV, López MR, de Vergara BC, et al. Spinal epidural hematoma in hemophilic children: controversies in management. Childs Nerv Syst 2009;25:987-91; discussion 993, 995.
42. Hosoki K, Kumamoto T, Kasai Y, et al. Infantile spontaneous spinal epidural hematoma. Pediatr Int 2010;52:507-8.
43. Johnstone RH, Leigh WB, Lawson EM, et al. Recurrent spontaneous spinal epidural haematoma treated with and without surgery. Injury Extra 2010;41:68-70.
44. Doymaz S, Schneider J. Spontaneous spinal epidural hematoma in a teenage boy with cholestasis: a case report. Pediatr Emerg Care 2013;29:227-9.
45. Lee CH, Song GS, Kim YH, et al. Spontaneous Spinal Epidural Hematoma Coexisting Guillan-Barré Syndrome in a Child: A Case Report. Korean J Spine 2016;13:167-9.
46. Mukai M, Miyagi M, Koyama T, et al. Spontaneous Spinal Epidural Hematoma in an Infant with Developmental Disabilities. Spine Surg Relat Res 2018;2:335-9.
47. Uei H, Tokuhashi Y, Maseda M, et al. Delayed-onset paralysis induced by spontaneous spinal epidural hematoma communicated with hematoma in the paraspinal muscle in a 6-month-old girl: a case report. Childs Nerv Syst 2019;35:379-83.
48. Morimoto D, Kim K, Kubota A, et al. Recurrent Cervical Spinal Epidural Hematoma: Case Report and Literature Review. NMC Case Rep J 2020;7:157-60.
49. Scalia G, Porzio M, Costanzo R, et al. Spontaneous spinal epidural hematoma in a pediatric hemophiliac. Surg Neurol Int 2023;14:69.
50. Moiyadi AV, Bhat DI, Devi BI, et al. Spinal epidural epitheloid hemangioma--case report and review of the literature. Pediatr Neurosurg 2005;41:155-7.
51. Pai SB, Maiya PP. Spontaneous spinal epidural hematoma in a toddler--a case report. Childs Nerv Syst 2006;22:526-9.
52. Lim JJ, Yoon SH, Cho KH, et al. Spontaneous spinal epidural hematoma in an infant : a case report and review of the literature. J Korean Neurosurg Soc 2008;44:84-7.
53. Sandvik U, Svensdotter E, Gustavsson B. Spinal cavernous extradural angiolipoma manifesting as a spontaneous spinal epidural hematoma in a child. Childs Nerv Syst 2015;31:1223-6.
54. Sano H, Satomi K, Hirano J. Recurrent idiopathic epidural hematoma: a case report. J Orthop Sci 2004;9:625-8.
55. Poonai N, Rieder MJ, Ranger A. Spontaneous spinal epidural hematoma in an 11-month-old girl. Pediatr Neurosurg 2007;43:121-4.
56. Iizuka Y, Iizuka H, Nagano K, et al. Laminoplasty preserving the deep extensor musculature for a pediatric cervicothoracic epidural hematoma case of hemophilia A. J Pediatr Orthop B 2014;23:584-8.
57. Sivakumaran R, King A, Bodi I, et al. Spontaneous epidural spinal haematoma in children caused by vascular

malformations. Eur Spine J 2016;25:614-8.
58. Ayres Pereira Harry Leite MIB, Alves SAF, Reinas RPV, et al. Sudden Onset of Severe Cervical Pain in an Adolescent Girl: Case Report and Review of Literature. Pediatr Emerg Care 2019;35:e248-e251.
59. de Amoreira Gepp R, Quiroga MRS. Recurrent spontaneous spinal epidural hematoma in children: case report. Archives of Pediatric Neurosurgery 2019;1:2-7.
60. Tailor J, Dunn IF, Smith E. Conservative treatment of spontaneous spinal epidural hematoma associated with oral anticoagulant therapy in a child. Childs Nerv Syst 2006;22:1643-5.
61. Duffill J, Sparrow OC, Millar J, et al. Can spontaneous spinal epidural haematoma be managed safely without operation? A report of four cases. J Neurol Neurosurg Psychiatry 2000;69:816-9.
62. Varsha J, Prashant S, Manohar S, et al. Spinal epidural hematoma in hemophilia-A a case report. Rivista di Neuroradiologia 2000;13:635-8.
63. Tamura K, Kanazawa T, Suzuki M, et al. Successful induction of immune tolerance by continuous infusion of recombinant factor VIII in a haemophilia A patient with high-inhibitor titres. Haemophilia 2006;12:100-2.
64. Heer JS, Enriquez EG, Carter AJ. Spinal epidural hematoma as first presentation of hemophilia A. J Emerg Med 2008;34:159-62.
65. Rathi N, Rathi A. Haemophilia A presenting as spontaneous spinal epidural haematoma in a 3-month-old infant. BMJ Case Rep 2010;2010:bcr0520102997.
66. Brown T, Purves E, Sargent M, et al. Clinical application of spinal ultrasound in screening for spinal haematoma in haemophilia A infants. Haemophilia 2011;17:318-20.
67. Per H, Canpolat M, Tümtürk A, et al. Different etiologies of acquired torticollis in childhood. Childs Nerv Syst 2014;30:431-40.
68. Thora A, Bhatnagar N, Tiwari A, et al. Nonoperative management of spontaneous epidural hematoma in hemophilia A. Int J Res Orthop 2017;3:202-6.
69. Acharya A, Bind R, Grewal S, et al. Spontaneous spinal epidural hematoma in a known case of hemophilia a managed conservatively: A rare case report. Journal of Applied Hematology 2021;12:112-4.
70. Villarreal-Martínez L, Sepúlveda-Orozco MDC, García-Viera DA, et al. Spinal epidural hematoma in a child with hemophilia A with high titer inhibitors and follow-up with prophylactic emicizumab: case report and literature review. Blood Coagul Fibrinolysis 2021;32:418-22.
71. Balkan C, Kavakli K, Karapinar D. Spinal epidural haematoma in a patient with haemophilia B. Haemophilia 2006;12:437-40.
72. Bisson EF, Dumont T, Tranmer B. Spontaneous spinal epidural hematoma in a child with hemophilia B. Can J Neurol Sci 2007;34:488-90.
73. Borkar SA, Prasad GL, Satyarthee GD, et al. Spontaneous spinal extradural hematoma in a child with hemophilia B, surgery or medical management - A dilemma? J Pediatr Neurosci 2011;6:131-3.
74. Nirupam N, Pemde H, Chandra J. Spinal epidural hematoma in a patient with hemophilia B presenting as acute abdomen. Indian J Hematol Blood Transfus 2014;30:54-6.
75. Kitagawa RS, Mawad ME, Whitehead WE, et al. Paraspinal arteriovenous malformations in children. J Neurosurg Pediatr 2009;3:425-8.
76. Cabral AJ, Barros A, Aveiro C, et al. Spontaneous spinal epidural haematoma due to arteriovenous malformation in a child. BMJ Case Rep 2011;2011:bcr0220113875.
77. Niimi Y, Sato S, Ryu B, et al. Unusual pediatric epidural arteriovenous fistula with venous ectasia presented with spontaneous spinal epidural hematoma: Case report. Interv Neuroradiol 2021;27:763-9.
78. Lee BJ, Jeong JH. Early Decompression in Acute Spinal Cord Injury : Review and Update. J Korean Neurosurg Soc 2023;66:6-11.
79. Li Y, Walker CL, Zhang YP, et al. Surgical decompression in acute spinal cord injury: A review of clinical evidence, animal model studies, and potential future directions of investigation. Front Biol (Beijing) 2014;9:127-36.
80. Zhang S, Geng F, Wang J, et al. Rapid Recovery of Spontaneous Spinal Epidural Hematoma without Surgical Treatment: Case Report and Literature Review. World Neurosurg 2018;115:216-9.

Cite this article as: Tombeng MA, Doi K, Mahadewa TGB, Tani S, Mizuno J. Spontaneous spinal epidural hematoma in children: a case report and literature review. J Spine Surg 2024;10(3):606-615. doi: 10.21037/jss-24-49