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CASE REPORT

Spontaneous thoracic epidural hematoma: a case report and literature review

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Abstract Spinal epidural hematoma is a rare neurosurgical emergency in respect of motor and sensory loss. Identifiable reasons for spontaneous hemorrhage are vascular malformations and hemophilias. We presented a case of spontaneous epidural hematoma in an 18-year-old female patient who had motor and sensory deficits that had been present for 1 day. On MRI, there was spinal epidural hematoma posterior to the T2–T3 spinal cord. The hematoma was evacuated with T2 hemilaminectomy and T3 laminectomy. Patient recovered immediately after the surgery. Literature review depicted 112 pediatric cases (including the presented one) of spinal epidural hematoma. The female/male ratio is 1.1:2. Average age at presentation is 7.09 years. Clinical presentations include loss of strength, sensory disturbance, bowel and bladder disturbances, neck pain, back pain, leg pain, abdominal pain, meningismus, respiratory difficulty, irritability, gait instability, and torticollis. Most common spinal level was cervicothoracic spine. Time interval from symptom onset to clinical diagnosis varied from immediate to 18 months. Spinal epidural hematoma happened spontaneously in 71.8 % of the cases, and hemophilia was the leading disorder (58 %) in the cases with a definable disorder. Partial or complete recovery is possible after surgical interventions and factor supplementations.

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Introduction

Spinal epidural hematoma (SEH) is a rare neurosurgical emergency in respect of motor and sensory loss. SEH occurring without a trauma is called as spontaneous SEH (SSEH). Identifiable reasons for spontaneous hemorrhage are vascular malformations and bleeding disorders. Incidence of SSEH is 0.1/100,000, and SSEH is common in fourth and fifth decades of life [1, 2]. Prompt diagnosis is very important for timely intervention in children [3].

We present an 18-year-old female patient having SSEH and discuss the literature in respect of prevalence, diagnostic tools, treatment approaches, and outcomes of SSEH in pediatric patients.

Case report

An 18-year-old girl was admitted to our clinic for difficulty in walking and sensory disturbances that had developed 1 day ago. In her neurologic exam, she was alert and oriented. She was paraparetic (strength of lower extremities=2/5), and her sensory level was at T3 dermatome. Her deep tendon reflexes were normoactive, and Babinski was negative, bilaterally. In her medical history, there was no recent trauma, no familial bleeding disorder, or no anticoagulation treatment. On MRI, posterior to the spinal cord, there was a mass lesion in the epidural space at T2–T3 levels, which was isointense on T1-weighted images and hypointense on T2-weighted images compared to cerebrospinal fluid intensity, consistent with acute hematoma (Fig. 1a, b). She was taken to surgery after

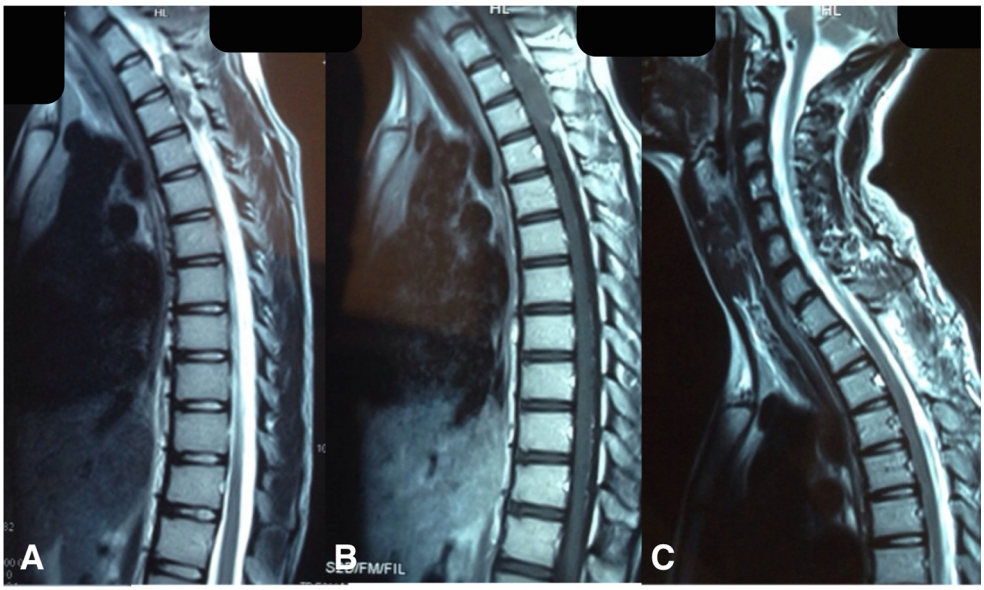

Fig. 1 Preoperative T2-weighted MRI depicts hypointense (**a**), and T1-weighted MRI depicts isointense posterior spinal epidural mass lesion, which is compatible with acute hematoma (**b**). No residual hematoma or myelopathy is present in postoperative T2-weighted MRI (**c**)

immediate clinical and laboratory evaluations had been completed. T2 hemilaminectomy and T3 laminectomy were performed in the operation. A blood clot was observed and was aspirated completely. Upper and lower spinal levels were clear for any additional presence of hematoma mass. No identifiable vascular malformation was noticed during decompression of the epidural space. Surgery was uneventful, and she recovered completely after the operation (Fig. 1c).

Discussion

Spinal hematomas are categorized into four groups: subdural, epidural, subarachnoid, and intramedullary. Spinal hematomas could lead to devastating results such as neurologic deficits and even death [3]. Spinal epidural hematoma, which is the most common type of spinal hematomas, presents rarely in pediatric population [2, 3]. Although there is confusion in definition of SSEH, we determined SEH that occurs without any trauma as SSEH. Identifiable underlying disorders beside trauma are spinal interventions, bleeding disorders, spinal infections, spinal tumors, and spinal vascular malformations [2, 4].

Spinal epidural hematoma in pediatric population was first described by Cooper in 1832 [5]. Literature review depicted 112 pediatric cases (including the presented one) of spinal epidural hematoma. Of these 112 patients, 70 were male and 39 were female (F:M=1.1:2, sex was not mentioned in three cases). Average age at presentation was 7.09 years (range=0–18 years). Clinical presentations include loss of strength, sensory disturbance, bowel and bladder disturbances, neck pain, back pain, leg pain, abdominal pain, meningismus, respiratory difficulty, irritability, gait instability, and torticollis. Clinical diagnosis is hard to make in infants, who have usually presented with non-specific symptoms such as irritability [3]. Most common spinal level was cervicothoracic spine (46.3 %). Other sites were thoracic (20 %), cervical (15.4 %), thoracolumbar (8.1 %), cervicolumbar (6.3 %), and lumbar (3.6 %). Time interval from symptom onset to clinical diagnosis varied from immediate to 18 months. Spinal epidural hematoma happened spontaneously in 71.8 % of the cases. Hemophilia was the leading disorder (58 %) in the cases with a known disease. Partial or complete recovery is possible after surgical interventions and factor supplementations (Table 1) [3, 5–73].

Exact pathogenesis of SSEH is not clear, yet. Spinal epidural venous plexus has been accused of bleeding source by many authors [3]. Spinal epidural venous plexus has no valves, and a sudden increase in pressure due to straining, voiding, crying, and coughing could lead to backflow of blood in the plexus and rupture of the venous vessels [2, 16, 40, 74]. There are infectious, inflammatory, and metastatic diseases in differential of SEH. Magnetic resonance imaging is superior to other diagnostic modalities in delineating location, consistency, and duration of the hematoma. Status of the spinal cord and any underlying pathology such as vascular malformations could also be evaluated with MRI [3]. In case of vascular malformations, additional imaging with angiography is necessary to better delineate the feeding and draining vessels.

Surgical decompression is the first-line treatment modality for SSEH [3, 40, 75, 76]. Laminectomy is the most effective decompressive approach in SSEH, yet it has some conflicts in pediatric patients due to progressive kyphotic deformity in

Table 1 Pediatric spinal epidural cases in the literature

Author(s)/year	Age/sex	Location	Etiology	Delay to treatment	Follow-up
Cooper/1832	12 years/M	C1–S	T	N/A	D
Abercrombie/1843	14 years/M	C2–C3	T	N/A	D
Jackson/1869	14 years/F	C1–C7	S	3 days	D
Johnston/1938	5 years/M	T8–T12	S (V)	1 year	D
Shenkin et al./1945	20 months/F	T1–T7	N/A	3 weeks	R
Carrea et al./1954	4 months/M	T9–L2	S	N/A	NR
Nichols and Manganiello/1956	15 years/M	C6–T1	S	N/A	R
Jones and Knighton/1956	12 years/M	C6–T2	S (H)	12 h	D
Lepintre et al./1956	8 years/F	C7–T1	S	N/A	R
Douglas and McAlpine/1956	16 years/M	Thoracolumbar	T	3 days	NR
Maxwell and Puletti/1957	4 years/M	T2–T4	S	Acute	R
Lepintre et al./1957	6 years/M	C7–T2	T	N/A	NR
Scott/1958	9 years/F	T1–T4	S	6 months	R
Lepoire et al./1961	15 years/M	C6–T1	N/A	N/A	R
Odom/1962	17 years/M	C5–C7	N/A	N/A	PR
Jackson/1963	14 months/F	T1–T5	T	N/A	R
Dawson/1963	15 years/M	L2–L5	S	N/A	R
Mayer/1963	17 years/F	C7–T1	S (V)	18 months	PR
Rao et al./1966	17 years/M	L4–L5	T	5 days	R
Rebello and Dastur/1966	11 years/M	C3–C7	T	Acute	PR
Cooper/1967	14 years/F	C3–C5	T	3 h	R
Hehman and Norrell/1968	21 months/M	C3–T9	T	24 days	PR
Posnikoff/1968	30 months/F	C5–T5	S	11 days	R
Amyot et al./1969	16 months/F	C5–C7	T	Weeks	R
Lepintre et al./1969	18 months/F	C3–C7	S	7 days	R
Pendl et al./1971	13 years/M	L5–S1	T	8 weeks	R
Nehlil et al./1972	17 years/M	C7	T	3 weeks	N
Pear/1972	15 years/M	T5–T7	T	N/A	R
Valladeres/1972	30 months/M	T3–T5	S	12 days	R
Tsai et al./1975	17 years/M	L3–L4	S	N/A	R
Grollmus and Hoff/1975	15 years/M	C6–T1	S	12 h	R
	33 months/F	C7–T3	T	2 days	PR
Packer and Cummins/1977	13 years/F	Upper thoracic	S	N/A	R
	17 years/M	T4	S	N/A	NR
Ghanem and Ivan/1978 and Ventureya et al./1979	8 years/M	C7–T2	T	4.5 days	R
Robertson et al./1979	6 years/F	T1–T3	S	N/A	PR
Gosnold and Sivaloganathan/1980	9 months/M	T6–T10	T	N/A	D
Vallee et al./1982	22 months/F	C5–T1	S	10 days	R
Wittebol and Van Veelen/1984	6 years/F	C5–T6	S	4 days	PR
Williams and Nelson/1987	7 years/F	C4–T2	S	72 h	PR
Matsumae et al./1987	13 years/M	C4–C7	S	2 days	PR
Pan et al./1988	13 years/F	C4–C7	T	4 h	R
Narawong et al./1988	4 years/M	T1–T5	S (H)	8 h	R
	8 months/M	C2–T3	S (H)	1 day	R
Calliauw et al./1988	13 years/M	Lower cervical–T9	T	2 days	R
Licata et al./1988	18 months/M	T1–T2	S	6 days	R
Nagel et al./1989	7 years/F	C4–C5	T	4 days	R
Epstein et al./1989	5 years/F	T3–T5	T	4 h	R
Faillace et al./1989	3 months	T8–L4	S (H)	N/A	NR

Table 1 (continued)

Author(s)/year	Age/sex	Location	Etiology	Delay to treatment	Follow-up
Jost et al./1990	7 years/M	Thoracolumbar	S (H)	N/A	R
Hamre and Haller/1992	9 years/M	C2–T6	S (H)	N/A	R
Tewari et al./1992	11 years/F	C5–C7	S	16 h	PR
	8 years/M	T10–L4	T	50 days	PR
	5 years/M	C5–T4	S	1 day	PR
Noth et al./1993	6 months	C2–T6	S (H)	N/A	R
Iguchi et al./1993	14 months/F	N/A	S	N/A	R
Joseph and Vinen/1993	17 years/F	C7–T2	S	2.5 days	PR
Sheikh and Abildgaard/1994	7 years/M	C3–L2	S (H)	N/A	R
Canderelli et al./1994	2 years/F	C5–T4	S	5 days	R
	16 months/F	C4–C7	S	1 week	R
Muhonen et al./1995	22 months/M	C5–T2	S (V)	5 days	R
Hutt et al./1996	11 months	T9–T12	S (H)	N/A	R
Patel and Garg/1996 and Patel et al./1998	4 years/M	C7–T1	T	4 days	R
	22 months/F	C7–T10	S	6 days	R
	18 months/M	Foramen magnum–T1	S	5 days	PR
Miyagi et al./1998	16 years/M	C2–C6	S (V)	N/A	R
Pecha et al./1998	13 years/M	T1–T4	S	8 h	PR
Alva/2000	10 months/M	C3–C6	T	5 days	R
Chrétiennot et al./2001	1 year/M	C7–L2	S (H)	2 days	R
	8 years/M	T11–L5	S (H)	1 day	R
Ravid et al./2002	10 years/F	C6–T1	S	9 days	R
Chang et al./2002	5 years/F	T6–T8	S	N/A	R
Chuang et al./2003	4 years/F	C4–T2	S (V)	10 days	PR
	4 years/M	C7–T3	S (V)	1 month	R
Liao et al./2004[a]	6 months/F	C7–T7	T	7 days	NR
	2 years/M	C2–T4	S	8 h	R
Iwamuro et al./2004	7 months/M	T1–L1	S (H)	9 days	R
Blount et al./2004	Neonate/M	C5–mid thoracic spine	S	No delay	NR
Tender and Awasthi/2004	12 years/F	N/A	S	2 days	R
Moiyadi et al./2005	5 years/M	C3–T4	S (V)	5 days	NR
Soundappan et al./2005	11 months/F	C6–T7	T	A few hours	R
Fountas et al./2006	12 years/F	C3–C7	T	A few hours	R
Watanabe/2006	4 years/F	T2–T4	T	Immediate	R
Pai and Maiya/2006	15 months/M	C4–T3	S	2 days	R
Balkan et al./2006	17 years/M	C6–T12, L5–S1 (minor)	S (H)	2 days	R
Tailor et al./2006	8 years/M	T12–L2	S	1 week	R
Poonai et al./2007	11 months/F	C4–T3	S	2 months	PR
Lee et al./2007	4 months/M	C4–T4	S	5 days	R
Güzel et al./2007	14 years/F	C4–T6	T	2 h	PR
Bisson et al./2007	7 years/M	C2–T3	S (H)	5 days	R
Lim et al./2008	20 months/F	C7–T4	S	14 days	R
Kalina et al./2008	7 months/M	C2–L4	S (H)	N/A	R
Ramelli et al./2008	7 months/F	C6–T7	S	2 weeks	NR
	13.5 years/M	C5–T2	S	N/A	PR
Kiran et al./2009	4 years/M	C7–T2	T	Immediate	PR
	4 years/M	T3–T6	T	Immediate	R
Kitagawa et al./2009	12 years/F	T1–T3	S (V)	6 months	PR
Lo/2010	11 years/M	T3–T5	S (V)	N/A	PR

Table 1 (continued)

Author(s)/year	Age/sex	Location	Etiology	Delay to treatment	Follow-up
	6 years/M	C7–T4	S (V)	2 days	PR
Hosoki et al./2010	11 months/M	T9–L1	S	3.5 days	PR
Kiehna et al./2010	5 months/M	C1–cauda equina	S (H)	1 month	R
Cabral et al./2011	9 years/M	C7–T4	S (V)	N/A	R
Min et al./2011	8 months/M	C7–T3	S (V)	40 days	R
Azumagawa et al./2012	12 years/M	C4–T4	S	5 days	R
Gupta et al./2012	6 years/M	C3–T4	S	A few hours	R
Schoonjans et al./2013	8 months/M	C5–L1	S	4 days	NR
Doymaz and Schneider/2013	14 years/M	C1–T2	S	1 day	PR
Paraskevopoulos et al./2013	8 years/M	C6–T2	S (V)	1 day	PR
Abbas et al./2013	5 months/M	C4–C7	S	N/A	N/A
Per et al./2014	5 years/M	C3–T2	S (H)	4 days	PR
Nirupam et al./2014	9 years/M	C6–T12	S (H)	5 days	D
Present case	18 years/F	T2–T3	S	1 day	R

F female, *M* male, *C* cervical, *T* thoracic, *L* lumbar, *S* sacrum, *DTR* deep tendon reflex, *N/A* not applicable, *S* spontaneous, *V* vascular malformation, *H* hemophilia, *T* trauma, *R* recovery, *PR* partial recovery, *NR* no recovery, *D* died

[a] Four more pediatric patients were described in the article, yet definitive information was not given in the text

upcoming years. For this reason, hemilaminectomy, laminotomy, and laminoplasty have been used in this patient population [3, 6–11, 14–16, 18–20, 22–24, 26, 28, 30–41, 45, 46, 48, 50, 52, 54–66, 77]. In our case, we preferred single-level laminectomy with single-level hemilaminectomy due to limited SSEH extension between T2 and T3 levels. In some cases with mild neurologic deficits and/or bleeding disorders, conservative approaches were preferred over surgical decompression [12, 13, 21, 23, 25, 27, 29, 42, 44, 49, 51, 53, 60, 61, 67–69]. The outcome of surgical interventions depends on preoperative neurological status of the patient, and the time interval passed from the onset of symptoms to surgery [3]. Critical deadline for timely intervention is 48 h for incomplete and 24 h for complete neurologic deficits [78]. The success rate of surgery is more profound in pediatric cases than adults, and even complete recovery has rarely been reported after late presentation [14, 40, 71, 72]. For this reason, delayed presentation and severe neurologic deficits are not contraindications for surgery if the patient does not have any underlying bleeding disorders [3, 5].

Conclusion

Spontaneous spinal epidural hematoma is a rare neurosurgical emergency, especially in pediatric population. Response to surgery (in non-coagulopathy situation) is devastating in this patient population despite delayed surgical intervention and severity of neurologic deficits. However, surgery should not be delayed, as soon as bleeding disorders have been eliminated from differential diagnosis list, for a better and fast recovery.

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Conflict of interest All the authors declare that there is no conflict of interest.

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