Case report

Spontaneous epidural hematoma of thoracic spine presenting as Brown-Séquard syndrome: report of a case with review of the literature

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Background: Spontaneous spinal epidural hematoma (SSEH) is an uncommon clinical entity. It produces a severe neurological deficit and prompt decompression is usually the first choice of treatment. Brown-Séquard syndrome is commonly seen in the setting of spinal trauma or an extramedullary spinal neoplasm, but rarely caused by SSEH.

Methods: Case report and literature review.

Findings: A previously healthy man presented with Brown-Séquard syndrome below T5–T6 cord segment secondary to spontaneous epidural hematoma. He opted for conservative treatment, which was followed by rapid resolution.

Conclusions: Although Brown-Séquard syndrome as a presenting feature of SSEH is rare, it does exist in exceptional case, which should be taken into consideration for differential diagnosis. Prompt surgical decompression is an absolute surgical indication widely accepted for patient with progressive neurological deficit. However, SSEH presenting with incomplete neurological insult such as Brown-Séquard syndrome might have a benign course. Successful non-operative management of this problem does not make it a standard of care, and surgical decompression remains the standard treatment for SSEH.

Keywords: Spinal epidural hematoma, Brown-Séquard syndrome, Thoracic vertebra, Spinal cord, Methylprednisolone, Paraparesis

Introduction

Spinal epidural hematoma is a rare pathology and could cause rapid development of quadriparesis/paraparesis, including a unique presentation as Brown-Séquard syndrome.¹ Although this bleeding event is frequently associated with hemorrhagic conditions, anticoagulants administration, or vascular malformation, it may also occur in a healthy subject as a spontaneous spinal epidural hematoma (SSEH). SSEH with neurological deterioration is usually treated early with surgical decompression.^{2,3} However, natural resolution with concomitant neurological improvement has also been reported in the literature.^{4–9}

This report describes a patient who sustained SSEH and presented with Brown-Séquard syndrome, which was followed by a rapid spontaneous resolution.

Case presentation

A previously healthy 35-year-old man suffered an insidious interscapular pain in the afternoon of the admittance day to the hospital. The pain worsened, becoming severe within a few hours before arriving to the emergency room of our hospital. Initial general examination was unremarkable, without history of fever, trauma, drug administration, or other suspicious systemic disease. Three hours later, the patient reported numbness of the right leg as well as weakness of dominant muscles in the left one. At that time, urinary retention was also noticed and catheter insertion was performed.

Neurological examination revealed paraparesis below T5–T6 spinal cord segment. Muscle strength on the left and right legs was 3/5 and 4+/5, respectively. Babinski reflex and ankle clonus were positive being more exaggerated on the right. Touch and tactile sensations were lost on both inferior limbs, but significantly on the left

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Figure 1 Sagittal (A) and transaxial (B) proton density images (3000 ms, TE 98 ms) 10 hours after the onset showed a hyperintense epidural mass at T3–T4 level. Hyperintense epidural mass extend through the T4/T5 intervertebral foramen (C, arrow).

one. Deficits in pain and thermal sensations were detected on the right leg.

Emergency magnetic resonance imaging (MRI) revealed an abnormal mass at T3–T4 level (Fig. 1A), occupying the left side of the spinal canal and compressing the spinal cord significantly to the contralateral side. The mass was hyperintense on both T2-weighted and proton density images (Fig. 1). According to the clinical and MRI findings, incomplete Brown-Séquard syndrome caused by SSEH was suspected.

Emergency decompression surgery was recommended. However, the patient and his family refused and opted for conservative treatment. A course with high dosage of steroids was instituted: an initial dosage of methylprednisolone (30 mg/kg, bolus injection) followed by a maintenance dosage (5 mg/kg/hour) for 3 straight days. Mannitol infusion was also prescribed simultaneously (20% mannitol, 250 ml intravenous infusion per 12 hours). Neurological examination was performed every hour to detect potential worsening of symptoms.

Symptoms improved dramatically within the next few hours. On the second day, approximately 15 hours after

the onset, the interscapular pain resolved and muscle strength of his legs improved to 5-/5 bilaterally. However, hyperactive Babinski reflex and ankle clonus remained positive in the right leg. Sensory disturbance improved on both sides, and bladder sphincter control returned to normal. On the third day of admission, the neurological deficit was completely resolved and the patient could walk with minimal orthotic support under supervision. MRI with contrast was taken on the 11th day after the onset, showing neither presence of epidural hematoma at T3–T4 nor other spinal abnormalities (Fig. 2). Two weeks later, the patient was discharged and returned to his daily activities. At 1 year follow-up, the patient remained asymptomatic without abnormal MRI findings.

Discussion

SSEH is a well recognized but rare condition, which may result in serious neurological insult to the spinal cord. It initiates with acute back or neck pain around the involved level and radiating to the corresponding dermatome, followed by a cord compression syndrome. Usually, the neurological deficits follow a course of

Figure 2 MRI with contrast 11 days later. T1-weighted MRI (A) and proton density image (B) demonstrated the resolution of the extradural hematoma. No other identifiable etiologies such as neoplasia, arteriovenous malformation were found.

rapid bilateral progression.¹⁰ Tetraparesis/paraparesis is commonly seen in such patients. However, as seen in this case, SSEH may cause asymmetric spinal cord compression causing an incomplete neurological deficit such as Brown-Séquard syndrome.

Brown-Séquard syndrome is characterized by ipsilateral loss of motor function resulting from corticospinal tract compression, combined with contralateral loss of pain and temperature sensations as a result of spinothalamic tract dysfunction.¹¹ It is most commonly seen following spinal trauma or extramedullary spinal neoplasm. Spinal epidural hematoma has rarely been considered to be a cause of Brown-Séquard syndrome. Kreppel *et al.*² reviewed 623 cases from 1826 to 1996 and found only 17 patients who sustained Brown-Séquard syndrome secondary to spinal epidural hematoma. Since then, other nine cases have been added to the medical literature.^{5,11–18} From the total number of 26 cases, all cases associated with coagulopathy, vascular malformation, or other sources of hemorrhage were excluded. The remaining 16 cases (including the present one) were reviewed for detailed neurological descriptions (Table 1).^{5,11–24}

In these 16 cases, the age at presentation ranged from 21 to 80 years with an average of 57.9 years. The most commonly involved region was the cervicothoracic spine. Thirteen patients received surgical decompression and laminectomy, most of them with satisfactory outcomes.

Spinal epidural hematoma usually appears in the cervicothoracic and thoracolumbar regions.¹⁰ These levels corresponded closely to the areas of profuse vasculature, especially the radicular branches of the vertebral arteries at C5–T2 and Adamkiewicz's artery at T9–T12.² The mid-thoracic spine (T3–T9) is rarely affected by spinal epidural hematoma; however, when it does occur at these levels, the neurological deficit is often complete. This is due to the much narrower spinal canal between T3 and T9 and a much smaller interpeduncular space than in the cervical or lumbar spine.²⁵ To our knowledge, this is the first report of a SSEH presenting with Brown-Séquard syndrome located at mid-thoracic spine.

Surgery is the first choice of the treatment for SSEH. It has been suggested that the earlier the surgery is performed after clinical onset, the better the prognosis. According to an extensive review by Groen and van

Table 1 Summary of 16 cases of SSEH presenting with Brown-Séquard syndrome

	First author/year	Age (year)/sex	Location of SSEH	Treatment	Outcome
1	Russman/1971 ¹⁹	53/F	C6	Decompression	Full recovery
2	Murata/1984 ²⁰	75/M	C2–C5	Decompression	Full recovery
3	Zupruk/1989 ²¹	80/M	Cervical spine	Decompression	Full recovery
4	Moran/1990 ²²	56/M	C5–C6	Decompression	Good recovery
5	Egido/1992 ²³	Aged/M	NR	Conservative management	Full recovery
6	Mahieu/1994 ²⁴	26/F	NR	Decompression	Full recovery
7	Ehsan/1996 ¹⁸	NR/M	NR	Decompression	Full recovery
8	Hancock/1997 ¹²	51/F	C5–T2	Decompression	Full recovery
9	Narberhaus/2002 ⁵	69/M	C6–T2	Conservative management	Full recovery
10	Adamson/2004 ¹³	63/M	C5	Decompression	Good recovery
11	Gill/2004 ¹¹	71/F	C5–C7	Decompression	Partial recovery
12	Spengos/2005 ¹⁵	66/F	C4–C7	Decompression	Full recovery
13	Seet/2005 ¹⁴	51/F	T10–T12	Decompression	No recovery
14	Domenicucci/2007 ¹⁶	52/F	Cervical spine	Decompression	Full recovery
15	Ofluoglu/2009 ¹⁷	63/M	C4–C6	Decompression	Full recovery
16	Present case	35/M	T3–T4	Conservative management	Full recovery

M, male; F, female; NR, no report; SSEH, spontaneous spinal epidural hematoma.

Alphen,²⁶ it was evident that prompt decompression (≤ 36 hours) was associated with favorable outcomes. On the other hand, the better the preoperative neurological status, the better the recovery postoperatively.

However, this patient refused surgery and had a rapid resolution of both clinical symptoms and radiographic findings with conservative treatment. Of previously reported cases (Table 1), only 3 of 16 cases (including the present one) resolved spontaneously. Comparisons of the results of operative treatment vs. conservative management (Table 2) showed that conservative treatment was not necessarily associated with severe neurological deficit. Nevertheless, the time window of spontaneous resolution is varied. Matsumura *et al.*²⁷ reported a hematoma in a patient that did not disappear even at 9 months from the time of onset. We presumed that this discrepancy of times might be explained by different origins and locations of bleeding. While some many authors assumed that SSEH was induced by rupture of the epidural veins, others advocated the arterial origin of bleeding, citing that the intrathecal pressure was higher than venous epidural pressure. If it is true that a different origin and location of bleeding may explain the discrepancy, the exact origin of bleeding in SSEH is still controversial.

The exact reason for spontaneous resolution is also still unknown. Serizawa *et al.*²⁸ suggested that a small and swift longitudinal spreading hematoma might relieve the pressure on the spinal cord through spreading a larger area leading to a spontaneous resolution. Inamasu *et al.*²⁹ considered the leakage of the liquid hematoma through the intervertebral foramen as a possible mechanism for the spontaneous decompression of the spinal neural structures. In the present case, SSEH extended through the T3–T4 and T4–T5 intervertebral foramina on MRI (Fig. 1C). Based on these observations, the leakage through these intervertebral foramina might have been the reason for the unexpected rapid resolution in this case.

The use of steroids and mannitol is a well-known measure for preventing the secondary injury to the spinal cord in clinical practice. However, a recent study by Ito *et al.* showed that no evidence supports the opinion that high-dose methylprednisolone sodium succinate administration facilitates neurological improvement in patients with spinal cord injury.³⁰ Kreppel *et al.*'s study² also showed that no specific treatment was performed in majority of patients who chose non-operative treatment. In SSEH, the primary spinal cord injury caused by mechanical compression is dominant; so steroids and mannitol might not be necessary or significantly impact the final outcome. However, its use may reduce the secondary injury to the spinal cord and give the surgeon a wider time window to perform the operation under the best possible conditions.

Table 2 Results of surgical decompression and conservative treatment

	Full recovery	Sequela	Total
Surgical decompression	11	2	13
Conservative treatment	3	0	3
Total	14	2	16

Conclusions

An uncommon clinical picture of Brown-Séquard syndrome caused by SSEH and rapid resolution occurred

in this 35-year-old man. Although Brown-Séquard syndrome as a presenting feature of SSEH is rare, it does exist and should be taken into consideration in the differential diagnosis. Prompt surgical decompression is an absolute surgical indication widely accepted for patients with progressive neurological deficit. However, SSEH presenting with incomplete neurological insult such as Brown-Séquard syndrome might have a benign course. For patients with a stable and improving neurological status, conservative management monitored by MRI might eventually be a treatment option, once a much more complete understanding of the pathophysiology of this disease is achieved in future. The successful non-operative management of this case is an insufficient evidence to consider it as a standard of care. Therefore, emergency decompression is still the standard treatment for SSEH.

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References

- Riaz S, Jiang H, Fox R, Lavoie M, Mahood JK. Spontaneous spinal epidural hematoma causing Brown-Sequard syndrome: case report and review of the literature. *J Emerg Med* 2007;33(3):241–4.
- Kreppel D, Antoniadis G, Seeling W. Spinal hematoma: a literature survey with meta-analysis of 613 patients. *Neurosurg Rev* 2003;26(1):1–49.
- Liu Z, Jiao Q, Xu J, Wang X, Li S, You C. Spontaneous spinal epidural hematoma: analysis of 23 cases. *Surg Neurol* 2008;69(3):253–60.
- Hentschel SJ, Woolfenden AR, Fairholm DJ. Resolution of spontaneous spinal epidural hematoma without surgery: report of two cases. *Spine* 2001;26(22):E525–E7.
- Narberhaus B, Rivas I, Vilalta J, Abos J, Ugarte A. Transient Brown-Sequard syndrome due to spontaneous spinal epidural hematoma. *Neurologia* 2002;17(7):384–7.
- 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(4):216–8.
- Schwarz SK, Wong CL, McDonald WN. Spontaneous recovery from a spinal epidural hematoma with atypical presentation in a nonagenarian. *Can J Anaesth* 2004;51(6):557–61.
- Schroder J, Palkovic S, Wassmann H. Spontaneous spinal epidural haematoma: a therapeutical challenge? Report of an unusual case. *Emerg Med J* 2005;22(5):387–8.
- Aoki Y, Yamagata M, Shimizu K, et al. An unusually rapid spontaneous recovery in a patient with spinal epidural hematoma. *J Emerg Med*, In press.
- Sanchez Rico P, Tomasa Irriguible TM, Catalan Eraso B, Martinez Vega S, Gener Raxach J, Klamburg IPJ. Acute spontaneous spinal epidural hematoma. *Med Intensiva* 2009;33(1):54–5.
- Gill D, Ifthikharuddin S, Samkoff LM. Acute Brown-Sequard syndrome. *Arch Neurol* 2004;61(1):131.
- Hancock JB, Field EM, Gadam R. Spinal epidural hematoma progressing to Brown-Sequard syndrome: report of a case. *J Emerg Med* 1997;15(3):309–12.
- Adamson DC, Bulsara K, Bronec PR. Spontaneous cervical epidural hematoma: case report and literature review. *Surg Neurol* 2004;62(2):156–159; discussion 159–160.
- Seet RC, Wilder-Smith EP, Ong BK, Lim EC. Spontaneous thoracic extradural haematoma presenting as the Brown-Sequard syndrome. *J Neurol* 2005;252(6):731–3.
- Spengos K, Sameli S, Tsvigoulis G, Vassilopoulou S, Vemmos K, Zakopoulos N. Spontaneous spinal epidural hematoma in an untreated hypertensive patient. *Eur J Intern Med* 2005;16(6):451–3.
- Domenicucci M, Ramieri A, Salvati M, Brogna C, Raco A. Cervicothoracic epidural hematoma after chiropractic spinal manipulation therapy. Case report and review of the literature. *J Neurosurg Spine* 2007;7(5):571–4.
- Ofluoglu E, Ozdemir A, Toplamaoglu H, Sofuoglu E. Spontaneous cervical epidural hematoma causing Brown-Sequard syndrome: case report. *Turk Neurosurg* 2009;19(1):99–102.
- Ehsan T, Henderson JM, Manepalli AN. Epidural hematoma producing Brown-Sequard syndrome: a case due to ruptured hemangioma with magnetic resonance imaging findings. *J Neuroimaging* 1996;6(1):62–3.
- Russman BS, Kazi KH. Spinal epidural hematoma and the Brown-Sequard syndrome. *Neurology* 1971;21(10):1066–8.
- Murata T, Ohhata K, Tsujikawa S, et al. A case of spontaneous spinal epidural hematoma presenting Brown-Sequard syndrome. *No Shinkei Geka* 1984;12(10):1195–200.
- Zupruk GM, Mehta Z. Brown-Sequard syndrome associated with posttraumatic cervical epidural hematoma: case report and review of the literature. *Neurosurgery* 1989;25(2):278–80.
- Moran M, Modravsky V, Fusek I. Spontaneous epidural hematoma with cervical localization-report of a case with etiopathologic analysis. *Psychiatr Neurol Med Psychol (Leipzig)* 1990;42(3):167–71.
- Egido Herrero JA, Saldana C, Jimenez A, Vazquez A, Varela de Seijas E, Mata P. Spontaneous cervical epidural hematoma with Brown-Sequard syndrome and spontaneous resolution. Case report. *J Neurosurg Sci* 1992;36(2):117–9.
- Mahieu X, Kridelka F, Pintiaux A, et al. Spontaneous cervical extradural hematoma in a pregnant woman. *J Gynecol Obstet Biol Reprod (Paris)* 1994;23(1):99–102.
- Groen RJ, Ponsen H. The spontaneous spinal epidural hematoma. A study of the etiology. *J Neurol Sci* 1990;98(2–3):121–38.
- Groen RJ, van Alphen HA. Operative treatment of spontaneous spinal epidural hematomas: a study of the factors determining postoperative outcome. *Neurosurgery* 1996;39(3):494–508; discussion 508–499.
- Matsumura A, Namikawa T, Hashimoto R, et al. Clinical management for spontaneous spinal epidural hematoma: diagnosis and treatment. *Spine J* 2008;8(3):534–7.
- Serizawa Y, Ohshiro K, Tanaka K, Tamaki S, Matsuura K, Uchiyama T. Spontaneous resolution of an acute spontaneous spinal epidural hematoma without neurological deficits. *Intern Med* 1995;34(10):992–4.
- Inamasu J, Hori S, Aoki K, Aikawa N, Maruiwa H, Toyama Y. Spontaneous spinal epidural hematoma. *Am J Emerg Med* 2000;18(7):837–9.
- Ito Y, Sugimoto Y, Tomioka M, Kai N, Tanaka M. Does high dose methylprednisolone sodium succinate really improve neurological status in patient with acute cervical cord injury?: a prospective study about neurological recovery and early complications. *Spine (Phila Pa 1976)* 2009;34(20):2121–4.