

CASE REPORT

The classification of recurrent spinal epidural hematoma: a review of the literature and a comparison with the cases

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Abstract Symptomatic postoperative spinal epidural hematoma (SEH) and spontaneous spinal epidural hematoma (SSEH) are both rare conditions, and recurrent SEH occurs even less frequently. Therefore, we describe a case of symptomatic postoperative SEH after surgical evacuation of SSEH, which was diagnosed using magnetic resonance imaging (MRI) and managed with negative pressure wound therapy (NPWT). The authors classified the reported recurrent SEHs into two types based on the cause of their previous hematoma, which can be classified as spontaneous or postoperative. The characteristics, diagnosis, managements, and results of recurrent SEHs were analyzed. The authors suggest that the postoperative SEH in the Type II will be treated with NPWT, and the new classification will be helpful for prognosis, diagnosis, and management of the recurrent SEHs.

Keywords Recurrent · Spinal epidural hematoma · Classification · Spontaneous · Negative pressure wound therapy

Introduction

Spinal epidural hematoma was first officially reported in 1869 by Jackson [1]. Since then, hundreds of SEHs have been reported. Conservative treatment and surgical

decompression are the common methods for SEHs. Surgical decompression includes laminectomy, hemilaminectomy, laminoplasty, interlaminar and endoscopic decompression [2]. Postoperative SEH usually occurs after surgical decompression. In this situation, the surgeon should make the original incision, evacuate blood clots, extend the laminectomy proximally or distally, and inspect the surgical site carefully [3]. Although patients with a SEH with ‘mild’ neurological deficits and without neurological deterioration in the early period may be treated conservatively, recurrent SEH as a special SEH should be treated more aggressively [4]. In the past, only 12 cases of recurrent SEH have been reported in the literature. Because of the rarity of the recurrent SEH, its characteristics, diagnosis and management remains unclear. We described a case of symptomatic postoperative SEH after surgical evacuation of SSEH, which was finally managed with NPWT. Moreover, we reviewed the literature, classified recurrent SEHs, and discussed the etiologies, characteristics, management strategies and outcomes based on the classification.

Case report

A 34-year-old male experienced sudden back pain combined with numbness and weakness in the lower limbs for 8 h with no trauma, drug use, family history or any disease history. Upon arrival to the emergency room, his blood pressure was 185/95 mmHg, and heart rate was 95 beats per min. On examination he had bilateral partial paralysis of the distal lower limb, anesthesia of bilateral lower limbs anesthesia below L4 and incontinence. The bilateral tendo Achillis reflexes and the bulbocavernosus reflexes were diminished. The Babinski’s sign was negative. Routine

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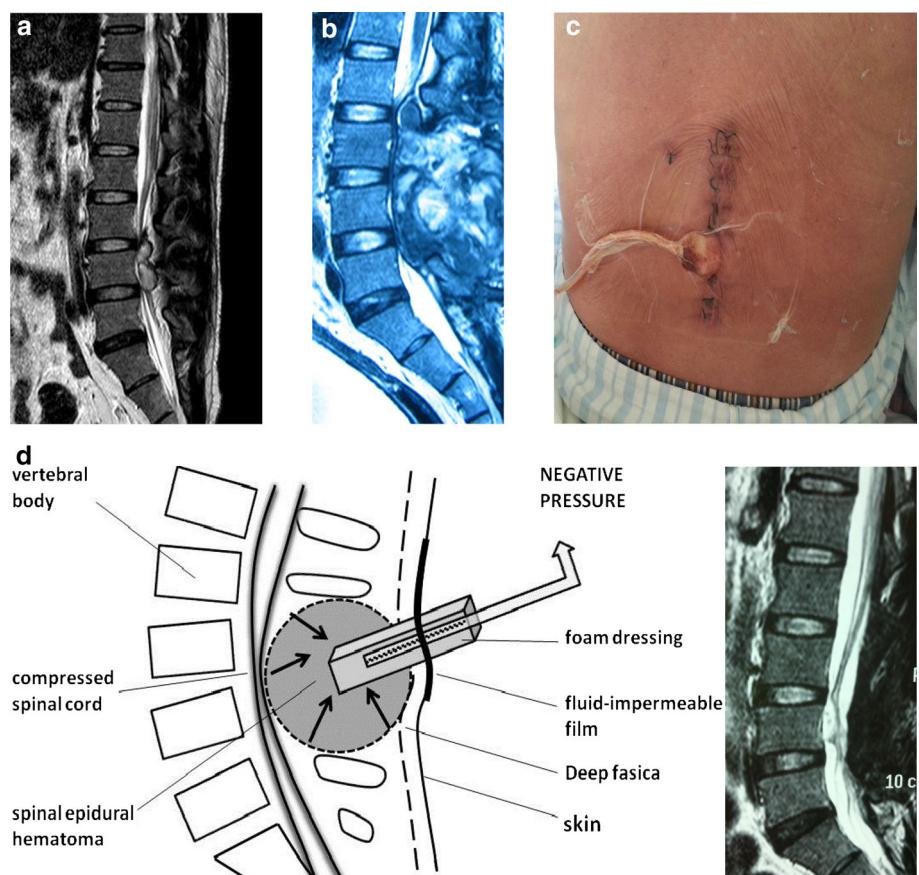
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laboratory tests indicated a mildly prolonged activated partial thromboplastin time of 42.3 s (normal 25–40 s). An emergent MRI showed a spinal epidural hematoma extending from L3 to L4 (Fig. 1a). The emergent test of coagulation factors activities showed a decreased coagulation factor VIII activity of 31.4 % (normal >75 %), and the patients were diagnosed with hemophilia A. After factor VIII replacement therapy (50 U/kg every 8 h) was performed to maintain it above 80 % of normal, the patient underwent an emergency laminectomy and hematoma evacuation (HE). Upon removal of the hematoma, he had nearly full recovery of muscle strength of his lower limbs. The intravenous dose of 50 U/kg of factor VIII every 8 h continued to maintain the factor VIII levels at 80 ~ 100 % after the operation.

However, on the eighth postoperative day the back pain, the patient had recurrent numbness and weakness in his lower limbs that rapidly worsened. His body temperature was 38.7 °C. Clinical examination demonstrated bilateral distal lower limbs paralysis and anesthesia, no deep tendon and pathological reflexes in both legs, and a decreased anal sphincter tone. Routine laboratory tests were within normal limits. MRI showed a huge hematoma compressing the dura and cauda equina severely (Fig. 1b). The patient was

diagnosed with symptomatic delayed postoperative SEH. After informed consent had been obtained, NPWT was performed with local anesthesia (Fig. 1c). A vacuum dressing [vacuum sealing drainage (VSD); Wuhan VSD Medical Science and Technology Co. Ltd., China] was used. Two or three stitches from the incision and deep fascia were removed, and the blood clots were evacuated appropriately. Active bleeding was not found and the laminectomy was not extended. A piece of polyvinyl alcohol foam was shaped to fit exactly within the wound. The foam wrapping around a suction tube was inserted into the center of the hematoma away from the dura using ultrasound guidance. The device and surrounding skin was sealed with a transparent adhesive membrane. The vacuum pressure was maintained at -125 mmHg (Fig. 1d). Systemic antibiotic with Cefuroxime was administered 3 days after surgery. His neurologic deficits improved immediately and recovered well after 24 h. The device was changed every 3 days with local anesthesia at the bedside and the treatment was applied for 5 days until ultrasound scan showed no remaining hematoma. Then, the incision and deep fascia was closed with two or three stitches. An intravenous dose of 50 U/kg of factor VIII every 8 h was continued to maintain the factor VIII levels at 80 ~ 100 %

Fig. 1 **a** Sagittal T2-weighted MR image revealed a posterior spinal epidural mass extending from L3 to L4. **b** Sagittal T2-weighted MR image demonstrated a huge hematoma compressing the spinal cord 8 days after the previous hematoma evacuation. **c** Negative pressure wound therapy was performed for the delayed postoperative spinal epidural hematoma. **d** Schematic of concept. **e** Sagittal T2-weighted MR image showed non apparent abnormality at 6 months follow-up



for 2 weeks. This dose was followed by 25 U/kg of factor VIII every 8–12 h to keep the factor VIII level above 50 %. The patient almost had no residual symptoms remaining except mild perineal numbness at 1-year follow-up (Fig. 1e).

Discussion

SSEH means spontaneous accumulating blood in spinal epidural space without obvious traumatic etiology such as a spinal fracture or an invasive spinal procedure. Symptomatic postoperative SEH after spine surgery is often presents as a serious complication causing neurologic impairment [5]. Recurrent SEH, which is less common, occurs after absorption or evacuation of the previous SEH [6, 7]. A PubMed review of the English literature for the terms “Spinal epidural hematoma” and “recurrent” yielded only 12 reported cases (Table 1). One case with recurrent paraplegia in a short time was excluded [8]. Recurrent SEHs can be divided into the following two types based on the cause of the last episodes: Type I is defined as recurring spontaneously after the previous SEH. Type II is defined as recurring SEH postoperatively after the previous SEH. Because no patient has had SSEH at a remote time from his or her previous spinal surgery before, the Type I disease undergoes SSEHs twice or several times. The Type II disease presents as SSEH and postoperative SEH twice or several times. Among the 13 patients (including our patient), 8 were diagnosed with Type I disease and 5 with Type II.

The Hernandez’s case with recurrent paraplegia and remission in 2 h was not diagnosed with recurrent SSEH but different periods of a SSEH [8]. Recurrent SEH was first reported in 1994 [9] and presents with symptoms of sudden severe spinal pain with or without any neurological deficits. The age of onset was from 6 to 65 years. The early detection of the compression status can be observed by MRI, which showed a lesion similar to that visualized during the first spontaneous or postoperative SEH. Currently, MRI is the preferred imaging modality for diagnosis of SEH.

There are several risk factors to SSEH, including hypertension, coagulopathy, anticoagulation therapy, thrombolytic medications, or spinal vascular malformations. However, predisposing factors in approximately 40–50 % of SSEHs remained unidentified [10]. In approximately 46.1 % of the recurrent SEHs the etiology was unidentified. The risk factors in recurrent SEHs are as follows: in the 13 cases (including our case), 2 patients took antiplatelet therapy with aspirin and clopidogrel, 1 case took antiplatelet therapy with aspirin, 3 had a coagulopathy, 1 suffered from vascular malformation, and 1 had

hypertension. Therefore, the etiology in recurrent SEHs is similar to the characteristics in SSEHs and postoperative SEHs [5].

Although the recurrent SEHs were very similar, there were still many differences. Therefore, we divided the cases into two types and found further similarities and differences. Of the 8 cases with Type I, 4 (50 %) presented in children younger than 18 years of age, 7 (87.5 %) occurred in the cervico-thoracic region, and only 2 (25 %) cases had a definite etiology. In the 5 cases with Type II, all the cases had existing identified factors and appeared in adult and older people, only 1 (20 %) occurred in the cervico-thoracic region. Although the causes of recurrent SEHs remained unclear, these data indicated that the Type I and Type II were associated with the identified etiology factors and onset location, respectively. The cervico-thoracic region is the most common location for SSEH [11, 22]. Therefore, most recurrent SSEHs cases with Type I disease also occurred in the cervico-thoracic region. The reason may be the maximum load at the cervico-thoracic region [6]. At this region (C7-T3), progression from cervical lordosis to thoracic kyphosis leads to transfer of weight from the posterior column to the anterior column and from relatively kinetic segments to stable segments that finally cause the local stress to increase. Moreover, most of the cases with Type II were associated with the identified etiology factors. Patients with the identified etiology inevitably presented with a high risk of surgical complications, as do the cases with postoperative Type II.

Conservative management of a SEH can be expected in a patient presenting with mild deficit or rapidly improving paralysis [4]. Emergency laminectomy and evacuation should be performed in patients with severe or worsening neurological signs within 8 h of the onset. Surgery later than 36 h is associated with a bad prognosis [11]. However, it is not easy to decide whether or not to perform the surgery because there is still no definite surgical indication for SEH. However, of 13 recurrent SEHs, 9 (69.2 %) had incompletely recovered at their last follow-up. Recurrent SEH is considered an indication for surgery at the time of the first recurrence [4, 6, 12]. It appears that recurrent compression, which is the second strike to the spinal cord, influences the recovery of the neurological condition. Therefore, patients with Type I should be treated with surgery at the time of the first recurrence. Type I disease, the recurrent SSEH, is caused by a extradural space-occupying lesion. As chronic SSEHs [13–17], sometimes it is very difficult to distinguish whether the lesion of Type I disease is a hematoma, tumor [13, 14, 17], abscess, extruded disc [14], or any other mass [16]. And MRI and histopathological examination are essential for the differential diagnosis. In addition to surgical treatment, we

Table 1 Literature review

Author/year	Age/sex	Etiology factor	Type	First episode (s)	Type of SEH	Symptom	Location	Therapy	Interval time			Last episode			Outcome
										Type of SEH	Symptom	Location	Therapy		
Franscini/1994 [10]	43 Y/M	AM	I	SSEH	SP	T10-12	CS	42 day	SSEH	SP	T10-12	L	CR		
Chen/1997 [26]	50 Y/M	Unknown	I	SSEH	SP and NS	T2-3	CS	30 day	SSEH	SP and NS	T2-3	L	ICR		
Sano/2004 [6]	6 Y/F	Unknown	I	SSEH	SP and NS	T1-3	CS	58 day	SSEH	SP and NS	T1-3	L	CR		
Abram/2007 [27]	10 Y/F	VM	I	SSEH	SP and NS	C7-T1	CS	1 year	SSEH	SP and NS	C6-T2	L	ICR		
Shin/2007 [28]	16 Y/F	Unknown	I	(1)SSEH	SP	C7-T2	CS	11 month	(3)SSEH	SP and NS	C7-T2	L	ICR		
Liao/2009 [29]	8 Y/F	Unknown	I	SSEH	SP and NS	C3-T8	CS	6 year	SSEH	SP and NS	C2-4	L	ICR		
Liao/2009	41 Y/M	C	II	SSEH	SP and NS	L1-5	L	48 h	POSEH	SP and NS	L1-5	E	ICR		
Liao/2009	53 Y/M	C, ESRD	II	SSEH	SP and NS	T4-L3	L	15 h	POSEH	SP and NS	T4-L3	E	ICR		
Lim/2011 [12]	57 Y/M	H, AM, CM	II	SSEH	SP and NS	T11-L3	L	1 day	POSEH	SP and NS	T11-L4	L	ICR		
Caruso/2012 [8]	65 Y/M	AM, CM	II	DPOSEH	SP and NS	C7-T1	L	17 day	DPOSEH	SP and NS	C7-T1	CS	ICR		
Jain/2014 [11]	20 Y/M	Unknown	I	(1)SSEH	SP and NS	T1-2	CS	30 day	(3)SSEH	SP and NS	T1-2	CS	CR		
Yamao [30]	39 Y/F	Unknown	I	(2)SSEH	SP	C6-T1	CS	4 month	(3)SSEH	SP	C6-T1	L	CR		
Current case	34 Y/M	C	II	SSEH	SP and NS	L3-4	L	8 day	DPOSEH	SP and NS	L3-4	NPWT	ICR		

AM aspirin medication, CM clopidogrel medication, VM vascular malformation, C coagulopathy, H hypertension, ESRD end stage renal disease, SSEH spontaneous spinal epidural hematoma, POSEH postoperative spinal epidural hematoma, DPOSEH delayed postoperative spinal epidural hematoma, SP spinal pain, NS neurological signs, CS conservative, CS incomplete recovery, E evacuation, NPWT negative pressure wound therapy, CR complete recovery, ICR Incomplete recovery

suggest NPWT, which is a minimally invasive approach that will manage the postoperative SEH in Type II cases.

The definition of NPWT was described explicitly by Fleischmann et al. in 1993 and developed further as a standard treatment in wound care in the 1990s [18–20], NPWT as an advanced wound therapy applies a persistent vacuum through a special sealed dressing and then draws out fluid and increases blood flow within the wound. Hematoma containing liquid and solid blood clots cannot be drained with ordinary drainage tube. However, the appearance of NPWT allowed us to drain the hematoma continuously without blocking the drainage tube. The use of this technique to remove the hematoma goes back to the very beginning when NPWT was first developed [19]. NPWT has been successfully applied in hematomas following high-energy trauma [21]. In postoperative SEHs surgical evacuation was essential until we made progress [22]. Therefore, we immediately chose NPWT to deal with our case with Type II disease and rapidly improved the neurological deficit. This technique will be performed without surgical decompression and extended laminectomy because of the sustained decompression by negative pressure. The advantages of NPWT in this situation are that it is safe and effective and does not require surgery. The disadvantage is longer treatment time than surgery. In addition, the device is removed after application for several days. Systemic antibiotic treatment is administered during the perioperative period. The mechanisms of promoting hematoma absorption by NPWT may be draining residual fluids and increasing wound blood circulation. At the last follow-up, the patient's neurological deficiency had nearly fully recovered except mild perineal numbness.

Hemophilia is a bleeding disorder that slows the blood clotting process. People with this condition experience prolonged bleeding or oozing following an injury, surgery, or having a tooth extraction. Hemophilia A can be divided into severe (<1 %), moderate (1–5 %), mild (>5 %) depending on the factor VIII activity [23]. When the factor activity reaches 25 %, patients show almost no coagulopathy [24]. Our patient had a factor VIII level of 1.4 %. Emergency decompressive laminectomy was the first choice for most SSEHs with severe and progressive symptoms [25]. However, early diagnosis of hemophilia A and appropriate treatment of the factor VIII replacement is essential.

Conclusion

Recurrent spinal pain with or without neurological deficits is the classical symptoms of recurrent SEH. MRI plays an important role in prompt diagnosis. Recurrent SEHs are divided into two types based on the cause of the last

episode, which can be classified as spontaneous or post-operative. The Type I disease often appears at the cervico-thoracic region and the Type II disease is associated with a known etiology. Patients with Type I should be treated with surgery at the time of the first recurrence. Patients with Type II disease can be treated by NPWT when the post-operative SEH occurs. We believe that the new classification will be helpful in prognosis, diagnosis and management of the recurrent SEHs.

Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

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