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General review

Spontaneous resolution and complete recovery of spontaneous cervical epidural hematoma: Report of two cases and literature review



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ABSTRACT

Objective. – To present the natural course and treatment modalities of spontaneous cervical epidural hematoma (SCEH), by reporting two rare cases with spontaneous resolution in both clinical and radiologic findings without surgery.

Material and methods. – One patient presenting with acute right side hemiparesis and another showing pure cervical radiculopathy were diagnosed with SCEH on magnetic resonance imaging (MRI). Both were both treated non-operatively. We also conducted a literature review of 19 cases of spontaneous spinal epidural hematoma (SSEH).

Results. – These two patients achieved complete resolution in terms of both neurologic function and radiologic findings within 21 days after onset. In the literature review, 63.2% of cases experienced neurologic improvement in the first 24 h, 78.9% achieved complete neurologic recovery within 1 month, and radiological images showed complete resolution of hematoma in the first month for 73.7% of patients.

Conclusions. – Atypical cervical SSEH can mimic cerebral stroke or a ruptured cervical disc. A high index of clinical suspicion followed by MRI examination is critical for diagnosis. Prompt surgical decompression and evacuation of the hematoma is generally regarded as first-line treatment. However, for patients without or with only slight neurologic symptoms, or showing early and sustained neurologic improvement, non-surgical therapy with close observation is a viable alternative. Both neurologic and radiologic resolution can be expected within the first month following onset in most cases of spontaneous resolution of SSEH.

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1. Introduction

Spontaneous spinal epidural hematoma (SSEH) is a rare emergent clinical condition that causes spinal cord compression. Annual incidence is 0.1 per 100,000 individuals [1]. SSEH usually presents with sudden onset of neck or back pain at the involved vertebral level, with radiating pain followed by rapidly progressive symptoms and signs of spinal cord compression [2,3]. It may lead to permanent neurological deficit or even death if diagnosis and treatment are delayed [4]. The clinical presentation can vary greatly when SSEH occurs in the cervical spine [5,6], and morbidity and mortality correlate strongly with cervical hematomas [7].

Prompt surgical decompression and evacuation of the hematoma is generally regarded as the optimal treatment for

SSEH, since symptom duration is reported to be associated with unfavorable outcome [2,7–9]. However, a handful of patients have also shown spontaneous resolution in terms of both clinical and radiologic findings even without surgery [10–25]. This makes selection of treatment modality difficult, given the complications liable to be induced by surgery. Here we report two uncommon cases of spontaneous cervical epidural hematoma (SCEH) that resolved under non-operative treatment, with complete neurologic recovery. The relevant literature was also reviewed, to determine the natural course of this disorder and appropriate management.

2. Case reports

2.1. Case 1

A 60-year-old man experienced sudden onset of severe neck and right shoulder pain during sleep, followed by progressive

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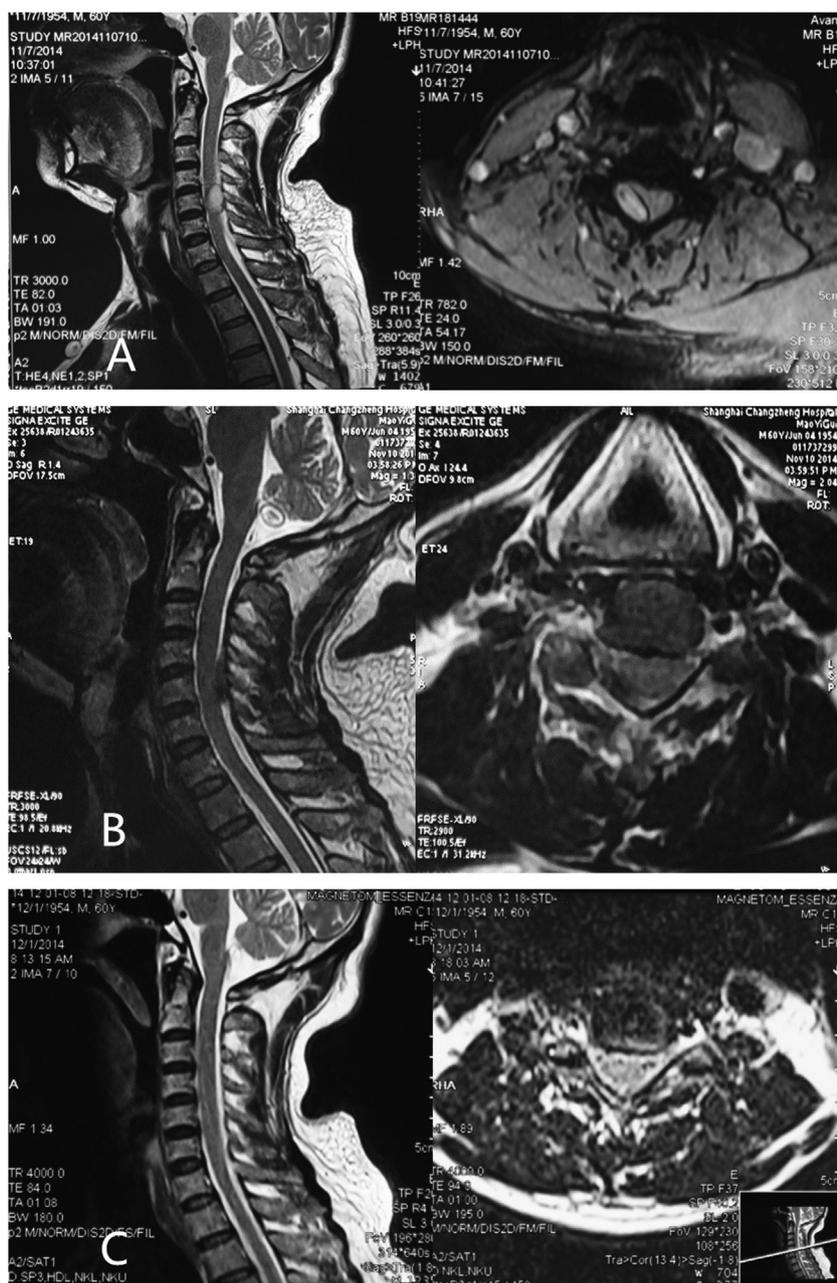


Fig. 1. A. Onset-MRI. Sagittal T2-weighted image (left) showing the epidural hematoma at C4–C6 level. It is hyperintense on T2-weighted image (left). Axial T2-weighted image (right) showing the epidural hematoma in the right posterolateral region compressing the right side of the spinal cord. B. Follow-up MRI (3 days). Size of the lesion has decreased observably. Intensity of the lesion has changed to slight hypointensity on T2-weighted images. C. Follow-up MRI (21 days). Sagittal (left) and axial (right) T2-weighted images showing complete disappearance of the epidural hematoma.

right-limb weakness. He was unable to stand and was brought to the emergency room 3 h after onset. Initial neurological examinations revealed intact consciousness, right side limb weakness (muscle power scored as 3/5 in the right upper and 2/5 in the right lower limb), and hypoesthesia at the C5 right dermatome. Right-limb deep tendon reflexes were absent. He was in good condition except for mild hypertension, with blood pressure at 145/94 mmHg on arrival. No history of trauma or anticoagulant therapy was reported. Blood tests, comprising platelet count, prothrombin time, partial thromboplastin time and biochemistry profile, were unremarkable. A computed tomography (CT) scan and diffusion magnetic resonance imaging (MRI) of the brain were taken to rule out intracranial hemorrhage or cerebral infarction. Cervical spine MRI revealed a well-defined ovoid lesion in the right posterolateral epidural region at C4–6 level, compressing the right side of

spinal cord. The lesion was isointense on T1-weighted and hyperintense on T2-weighted images (Fig. 1A). SCEH was suspected. No flow voids or post-contrast enhancement were present.

Intravenous dexamethasone was administered immediately, and emergency decompression surgery was planned. During preparation for surgery, pain and limb weakness unexpectedly started to improve spontaneously, 8 h after onset; right hemiparesis improved to grade 4/5. Surgery was therefore deferred, and non-operative treatment with close neurological and radiologic monitoring was implemented. Three days after admission, he was almost symptom-free except for mild neck pain and was able to walk without any assistance. Meanwhile, repeat MRI 3 days after onset showed decreased hematoma, and intensity on T1-weighted images had become heterogeneous with hyperintense areas, while T2-weighted images revealed slight hypointensity for

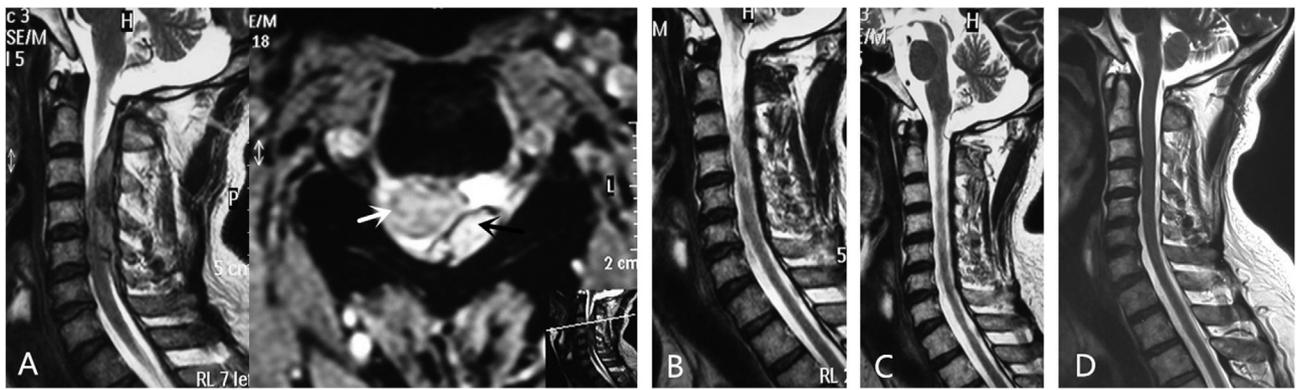


Fig. 2. A. Onset-MRI. Sagittal T2-weighted image (left) showing the epidural hematoma at C2–C6 level with heterogeneous intensity. Axial T2-weighted image (right) showing mass effect caused by the well-defined epidural hematoma (black arrow) creating rightward shift of the spinal cord (white arrow). Direct cord compression is not obvious and the spinal canal “buffer space” is relatively large. B. Follow-up MRI (7 days). Sagittal T2-weighted image showing observably decreased hematoma size. C. Follow-up MRI (21 days). Sagittal T2-weighted image showing complete disappearance of the epidural hematoma. D. Follow-up MRI (5 months). Sagittal T2-weighted image showing mild C3/4 disc herniation with no evidence of SSEH recurrence.

the lesion (Fig. 1B). The patient achieved complete neurological recovery 21 days after onset, with completely resolved SCEH on MRI images (Fig. 1C). There was no recurrence or neurological deficit during 1 year’s follow-up.

2.2. Case 2

A 53-year-old man was admitted with sudden onset neck pain radiating to the left shoulder and upper limb, accompanied by 1 day’s numbness and weakness in the left upper limb. Medical history was unremarkable, with no neck trauma, bleeding disorder or drug use. Physical examination revealed grade 3/5 weakness of major muscle groups in the left upper limb and decreased sensation to pin-prick and light touch between the left C4 and C6 dermatomes. Reflexes were normal. Based on clinical symptoms and signs, cervical spondylotic radiculopathy was suspected, and cervical MRI was performed. The result revealed a well-defined fusiform mass compatible with a hematoma in the left posterolateral epidural region, extending from C2 to C6 (Fig. 2A). Sagittal T2-weighted images showed mass with heterogeneous intensity. Axial T2-weighted images demonstrated the mass effect caused by the well-defined epidural hematoma, creating rightward shift of the spinal cord. Direct cord compression was not obvious on images, and the spinal canal space was relatively wide (Fig. 2A). Laboratory study results were all within normal limits.

Considering the presentation of pure cervical radiculopathy without signs or symptoms of cord compression, non-operative treatment was decided on, with close neurological and radiologic monitoring. Non-steroid anti-inflammatory drugs and intravenous dexamethasone were administered for symptomatic treatment, and a Philadelphia neck collar was used for neck immobilization. Numbness and weakness improved gradually. Seven days after onset, the patient was free of symptoms, and MRI showed observably decreased hematoma size (Fig. 2B). Twenty-one days after onset, complete resolution of epidural hematoma (Fig. 2C) was demonstrated on MRI, and the patient remained asymptomatic. At 5 months’ follow-up, the patient complained of occasional left shoulder pain without sensorimotor deficit, and repeat MRI showed mild C3/4 disc herniation with no evidence of SSEH recurrence (Fig. 2D).

3. Discussion

SSEH is a rare cause of spinal cord and radicular compression, accounting for less than 1% of all spinal epidural space-occupying lesions [26]. Though any vertebral segments may be involved,

location is predominantly in cervicothoracic and thoracolumbar dorsal areas [7,8], SSEH is most often attributed to spontaneous collection of blood in the spinal epidural space, without any traumatic or iatrogenic cause [8]. However, this does not exclude predisposing factors such as coagulopathy, vascular malformation, antiplatelet or anticoagulant therapy, cavernous angioma or tumor [11,27–30]. Other authors argue that only cases of idiopathic origin should be considered as SSEH [30]. Approximately 40 to 61% of cases without any underlying reported cause could match the criteria for idiopathic SSEH [31].

SSEH usually presents with sudden onset of neck or back pain at the involved vertebral level, with radiating pain followed by rapidly progressive symptoms and signs of spinal cord compression [2,3]. However, early accurate diagnosis is a challenge, especially in cervical SSEH (SCEH) [6]. Due to varying rapidity of onset and severity of upper spinal cord and radicular compression, various symptoms in SCEH can mimic cerebral stroke, ruptured cervical disc or cervical arterial dissection [10,32]. Therefore, MRI is critical for diagnosis of SCEH and can reveal the location and extent of the hematoma and the severity of spinal cord compression and spinal cord edema. In the present case No. 1, presentation with acute right side hemiparesis and history of hypertension easily led to an initial impression of cerebral stroke. Although the patient was diagnosed correctly, he could have been misdiagnosed as having cerebral infarction if cervical MRI had not been performed, and would have deteriorated severely if intravenous thrombolysis had therefore been attempted [6,33]. Interestingly, in case No. 2, pain, numbness and weakness in a single limb was the major presentation, which was consistent with radicular lesion. This is very rare in SCEH and has been reported only once [10]. On the axial T2-weighted image (Fig. 2A), the spinal canal “buffer space” was relatively large, and the mass effect of the well-defined epidural hematoma pushed spinal cord to the right, although direct cord compression was not clear. Hence we speculated that rightward spinal cord displacement might stretch the contralateral cervical nerve roots and cause radiculopathy, similar to the hypothesis of C5 nerve root palsy following cervical decompression surgery [34]. The experience derived from this case and the literatures implies that SCEH presenting isolated radicular involvement is more suitable for non-operative treatment [10,35].

We conducted a comprehensive search of the medical literature on PubMed, Embase and Web of Science in January 2018. Search terms were: (epidural hematoma) and (spontaneous) and (spin*). A total of 17 spontaneously resolved SSEH patients were identified in the English-language literature, with radiological imaging proving complete disappearance of the epidural hematoma. The characteristics and natural course of these patients as well as the

Table 1
Summary of reported SSEH cases with spontaneous resolution in both clinical and radiologic findings without surgery.

Author and year	Age (yrs), Gender	History	Level	Position	Radiology	AIS Grade	Why non-operative	Improvement interval	Neurologic recovery interval	Radiologic resolution interval
Present study	60, M	Hypertension	C4-C6	Dorsolateral	MRI	D	Spontaneously improved	8 h	21 d	21 d
Villas et al., 2006	53, M	No special	C2-C6	Dorsolateral	MRI	E	Mild symptoms	1 d	7 d	21 d
	64, M	Anticoagulant	C4-C5	Dorsolateral	MRI	E	Misdiagnosis	2 d	7 d	1 m
Subbiah et al., 2010	65, M	Anticoagulant	C4-T1	Ventral	MRI	C	Comorbidities	48 h	1 m	1 m
Silber et al., 1996	51, M	NSAID	C5-T4	Dorsolateral	MRI	B	Patient refusal	24 h	1 m	1 m
Sei et al., 1991	61, F	No special	C4-C7	Dorsolateral	MRI	Unknown	Spontaneously improved	5 d	24 d	18 d
Schroder et al., 2005	80, M	Hypertension	L1-L5	Ventral	MRI	D	Spontaneously improved	4 h	24 h	5 m
Sathirapanya et al., 2013	62, M	Hypertension	C7-T4	Ventral	MRI	A	Spontaneously improved	20 h	4.5 m	Unknown
Saito et al., 1994	49, M	No special	L2-L3	Dorsal	MRI	E	Patient refusal	1.5 m	2 m	10 m
Lemmens et al., 2012	66, F	No special	C2-C7	Dorsal	MRI	Unknown	Spontaneously improved	Hours	Hours	1 wk
Kato et al., 1994	54, M	Hypertension	C2-C7	Ventral	MRI	A	Spontaneously improved	8 h	10 d	1 m
Ishida et al., 2011	56, F	No special	C3-C5	Dorsolateral	MRI	Unknown	Mild symptoms	Unknown	Unknown	8 d
Hentschel et al., 2001	68, M	No special	C7-T1	Dorsal	MRI	B	Spontaneously improved	15 min	1 d	4 m
	65, F	Hypertension	T7-T12	Ventral	CT MRI	A	Spontaneously improved	3 h	1 m	3 m
Buyukkaya et al., 2014	46, M	No special	C5-T1	Dorsolateral	MRI	C	Spontaneously improved	Unknown	48 h	48 h
Aoki et al., 2012	64, M	Antiplatelet	C2-T3	Dorsal	MRI	A	Spontaneously improved	5 h	3 d	24 d
Anderson et al., 1989	63, F	Antiplatelet	C3-C7	Dorsolateral	CT	Unknown	No deterioration	24 h	3 m	3 wk
Wagner et al., 1996	74, M	Antiplatelet	C2-C6	Dorsolateral	MRI	Unknown	Spontaneously improved	12 h	13 d	3 d
Halim et al., 2008	25, M	No special	C2-T1	Ventral	MRI	A	Delayed diagnosis	10 d	24 d	11 d

AIS Grade: ASIA Impairment Scale Grade; CT: computed tomography; Improvement interval: time between initial onset and neurological improvement; MRI: magnetic resonance imaging; Neurologic recovery interval: time between onset and complete neurologic recovery; NSAID: nonsteroidal anti-inflammatory drugs; Radiologic resolution interval: time between onset and resolution of hematoma on radiologic findings.

two present cases are detailed in Table 1. Male patients were more likely to be treated non-operatively, accounting for 73.7% of spontaneously resolved patients. As hypertension was considered uncorrelated with the SSEH [36], anticoagulant or antiplatelet therapy was the most common predisposing factor (5/19 = 26.3%). A majority of these spontaneously resolved patients showed cervical hematoma (16/19 = 84.2%) and almost half had SCEH (9/19 = 47.4%). Five patients (5/19 = 26.3%) had complete neurologic deficit (ASIA impairment scale grade A) and 6 (6/19 = 31.6%) had incomplete neurologic deficit (ASIA grade B, C or D). Spontaneous improvement at or before medical evaluation was the most likely cause (12/19 = 63.2%) of selection for non-surgical therapy. Misdiagnosis, delayed diagnosis, associated comorbidities with high surgical risk or patient's refusal of surgery were other reported reasons for not operating. Neurological improvement time ranged from 15 min to 1.5 months, and 12 of the 19 patients (12/19 = 63.2%) experienced neurologic improvement in the first 24 h following onset. Neurologic recovery time ranged from 24 h to 4.5 months, and a majority of these spontaneously resolved patients (15/19 = 78.9%) showed complete recovery within 1 month of onset. Radiologic resolution time ranged from 3 days to 10 months, and MRI or CT imaging proved that 73.7% patients (14/19) showed complete resolution of epidural hematoma in the first month.

Nevertheless, prompt surgical decompression and evacuation of the hematoma is still the first-line option for SSEH [2,7–9]. Laminectomy or hemilaminectomy is the most common operative approach

[8], and laminoplasty is recommended for SCEH [2]. Several major case series documented surgical outcomes [2,3,8,9,31,37]. Prognosis is strongly dependent on initial neurologic status: patients with incomplete neurologic deficit have better outcomes than those with complete deficit [3,8,37]. In the case series reported by Liao et al., 88.9% of patients with incomplete neurological deficit achieved complete recovery within 1 year, versus only 37.5% of those with complete deficit [3]. Shorter interval between symptom onset and surgery was associated with better outcome [7,9], and even patients with complete neurologic deficit achieved better functional neurologic recovery when decompression was performed within 24 h [9]. Shorter progression interval between primary symptom onset and onset of neurological deficit and spinal cord edema on MRI [8,31] were also reported to be associated with poor prognosis. Therefore, due to the unpredictable course of SSEH, early surgical intervention before onset more serious neurological deficit is the generally accepted attitude in SSEH.

Therefore, in selected patients without or with only slight neurologic symptoms, or showing early sustained neurologic improvement, non-surgical therapy with close observation and repeated MRI follow-up is a viable treatment option that can achieve excellent neurologic outcome. However, considering that most spontaneously resolved SSEH patients experienced neurologic improvement in the first 24 h, if neurologic improvement is not observed within the first 24 h in a patient with severe neurologic deficit, or if neurologic deterioration occurs and neurologic

improvement stops at an unacceptable level, surgical intervention should be undertaken as soon as possible.

4. Conclusion

SCEH is a rare clinical emergency, needing prompt diagnosis and treatment. Atypical presentations can mimic cerebral stroke or ruptured cervical disc, and MRI examination is critical for diagnosis. For patients without or with only slight neurologic symptoms or showing early sustained neurologic improvement, non-surgical therapy with close observation and repeated MRI follow-up is a viable option that can achieve excellent neurologic outcome. However, for patients in whom neurological function is not improved within the first 24 h or deteriorates continuously, prompt surgical intervention should be undertaken.

Review board approval

All treatments performed in the study were in accordance with the ethical standards of the Ethics Committee of Shanghai Changzheng Hospital and the 1964 Helsinki Declaration and its later amendments, or comparable ethical standards.

Patient consent

Patients consented to the submission of this case report to the journal.

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Disclosure of interest

The authors declare that they have no competing interest.

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