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Case Report

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Recurrent Cervical Spontaneous Spinal Epidural Hematoma with Conservative Management: A Case Report

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Conflict of Interest
The authors have no financial conflicts of interest.

ABSTRACT

Cervical spontaneous spinal epidural hematoma (CSSEH) is a rare condition that can be potentially fatal if not properly diagnosed and managed. While prompt surgical decompression and evacuation of the hematoma are generally considered as the first line of treatment, mild cases that were managed through observation and conservative treatment have been reported. Our patient was a 24-year-old man who experienced two CSSEH events 8 months apart, both of which were managed conservatively. This was a rare case of recurrent CSSEH in which recovery was achieved without surgical intervention. We believe conservative treatment with close observation may be effective in CSSEH patients presenting with mild neurologic symptoms who have a tendency towards spontaneous neurologic improvement.

Keywords: Cervical spontaneous spinal epidural hematoma; Recurrent; Conservative management

INTRODUCTION

Cervical spontaneous spinal epidural hematoma (CSSEH) is a rare disease that can cause spinal cord compression, with an estimated incidence of 0.1 in 100,000 per year.[2] The typical signs and symptoms of CSSEH are acute neck pain accompanied by radiating pain at the associated level of the spine, and are rapidly progressive with signs of spinal cord compression.[2] These symptoms can lead to irreversible neurological deficits, such as paraplegia, quadriplegia, cardiac arrest, and death. Thus, CSSEH should be diagnosed and treated in an urgent manner.[6] Surgical treatment involving the decompression and evacuation of the hematoma is usually the initial treatment of choice.[12,16] However, some patients have also shown spontaneous resolution in terms of both clinical and radiologic findings without surgical intervention (**TABLE 1**).[1,3,5,7,11,13-15,17,19]

We report the case of a 24-year-old patient with recurrent CSSEH. This patient had sudden onset quadriparesis of motor grade 4 out of 5 in the first event, and 8 months later, left-sided hemiparesis of motor grade 4 out of 5 in the second event. Both CSSEHs were resolved with conservative management, with complete recovery of neurological deficits observed within a few days.

TABLE 1. Summary of patients with recurrent spontaneous spinal epidural hematoma

Study	Age (years) & sex	Medical history	Level of injury	Ventral hematoma?	Treatment (duration to recurrence)			Source of bleeding
					1st	2nd	3rd	
Harik (1971)[9]	20 M	-	T1-2	–	ob	ob	ob (3 months)	Not listed
Pear (1972)[15]	27 F	Pregnancy	C4-T1	–	op	ob (9 years)	-	Not listed
Matsumae (1987)[13]	8 F	-	1st; C2-4 2nd; C3-T8	–	op	op (6 years)	-	Engorged epidural vessels
Franscini (1994)[5]	50 M	-	T2-3	+	ob	ob (1 year)	op (1 month)	Not listed
Chen (1997)[3]	17 F	-	C7-T1	–	ob	ob (14 months)	op (4 months)	Angioma
Sano (2004)[17]	16 F	-	C7-T2	+	ob	op (5 months)	op (11 months)	Not listed
Groen (2004)[7]	10 F	-	C7-T1	–	ob	ob (2 months)	op (12 months)	Not listed
Abram (2007)[1]	13 M	-	C4-7	–	ob	op (38 months)	-	Not listed
Jain (2014)[11]	39 F	-	C6-T1	–	ob	ob (15 months)	op (4 months)	Venous plexus
Yamao (2015)[19]	6 F	-	T1-3	–	ob	op (2 months)	-	Not listed
Iwatsuki (2015)[10]	43 M	GPD	T10-12	–	ob	op (1.5 months)	-	Not listed
Morimoto (2020)[14]	13 F	-	C6-T1	+	ob	ob (2 months)	op (13 months)	Free epidural artery

C: cervical, GPD: partial platelet glycoprotein Ia/IIa deficiency, F: female, M: male, ob: observation therapy, op: operation pregnancy, T: thoracic.

Given that the presentation of CSSEH can vary greatly between patients, the choice between surgery or conservative treatment depends highly on individual factors. Here we summarize our individual case study in addition to previous literature of CSSEH cases to identify potential factors which may indicate conservative treatment in lieu of surgical intervention.

CASE REPORT

In July 2018, a 24-year-old male presented to the hospital with sudden-onset motor weakness in both legs following a fall down the stairs. There was no remarkable past medical history or familial history, including trauma. The patient was not taking any antiplatelet or anticoagulant drugs, and laboratory findings showed no signs of coagulopathy. He was not experiencing any constitutional symptoms, including fever, chills, or weight loss.

On examination, the patient had posterior neck pain with pain in both shoulders, and muscle power was scored as 4 out of 5 in both lower and upper limbs. Cervical spine magnetic resonance imaging (MRI) was performed, including contrast-enhanced MRI for differential diagnosis. The MRI revealed CSSEH with a characteristic biconvex-shape in cervical spine levels 4–6, which was compressing the left dorsolateral side of the spinal cord (**FIGURE 1**). The lesion was hypo- to iso-intense on T1-weighted, and hypo- to heterogeneous-intense on T2-weighted images. There was no evidence of tumorous or infectious lesions on contrast-enhanced images. Angiography was planned initially but was canceled due to a lack of

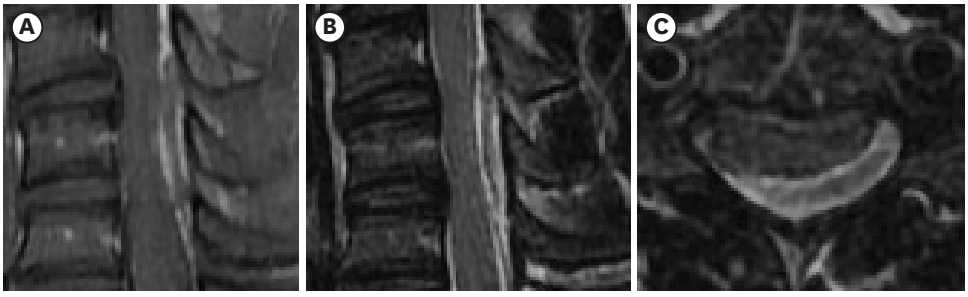

FIGURE 1. First cervical spontaneous spinal epidural hematoma event. Cervical-spine magnetic resonance imaging of the patient on July 19, 2018 showed a hypo-signal mass in cervical-spine levels 4–6 with spinal cord compression. (A) T1 contrast-enhanced, (B) T2 weighted sagittal view, (C) T2 weighted axial view, cervical-spine level 5.

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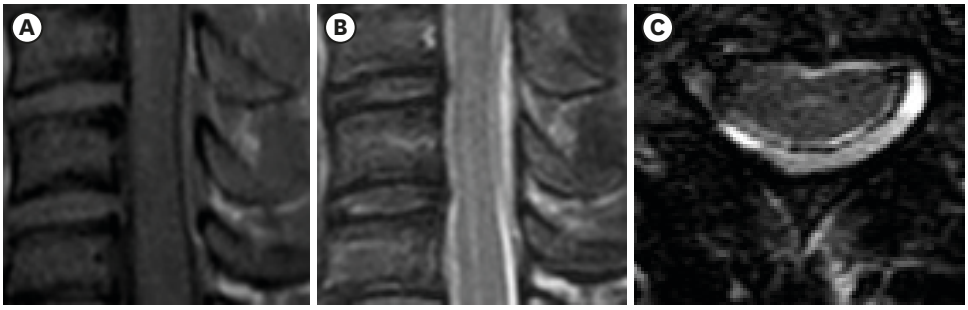

FIGURE 2. First cervical spontaneous spinal epidural hematoma event 3 month-follow up. A follow-up cervical-spine magnetic resonance imaging on October 25, 2018 showed hematoma regression in cervical-spine levels 4–6. (A) T1 contrast-enhanced, (B) T2 weighted sagittal view, (C) T2 weighted axial view, cervical-spine level 5.

evidence of vascular lesion-like flow voids. Since his symptoms had already improved at the time of arrival in the hospital, conservative management was planned in ambulatory care. A few days later, his leg motor power improved, and a follow-up MRI on October 25, 2018 indicated complete hematoma regression in cervical spine levels 4–6 (**FIGURE 2**).

On March 7, 2019, the patient returned to the hospital, experiencing sudden-onset paresthesia in all limbs, mild left-sided weakness with motor power scored at 4 out of 5, and mild gait disturbance. Cervical spine MRI was performed without contrast-enhanced imaging because it was too early to suspect tumor and vascular malformation from the last cervical spine MRI. The MRI revealed a 4.0 × 0.4 cm-sized mass in cervical spine levels 4–6, which was hypo-and heterogeneously intense on T2 images and was diagnosed as CSSEH (**FIGURE 3**). Because his motor weakness improved during admission, the patient refused to undergo surgery for CSSEH and was treated conservatively. His left side motor power improved to 5 out of 5 points, 2 days after admission, and paresthesia disappeared. He was discharged with complete neurological recovery and scheduled for follow-up in ambulatory care. On April 11, 2019, a follow-up MRI was performed, and a hematoma regression was confirmed (**FIGURE 4**).

DISCUSSION

Spinal epidural hematoma is a rare pathology associated with trauma, tumor, coagulopathy, vascular malformation, and idiopathic causes.[2] In contrast, spontaneous spinal epidural hematoma (SSEH) is a spinal epidural hematoma that occurs in the absence of any trauma, disease, or iatrogenic procedures with an incidence of 0.1 in 100,000 per year.[2,6] With rare

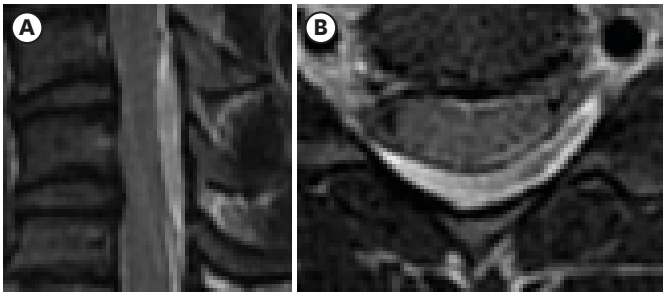

FIGURE 3. Second cervical spontaneous spinal epidural hematoma event. Cervical-spine magnetic resonance imaging on March 7, 2019 revealed a hypo-intensity mass in C-spine 4-5-6 levels. (A) T2 weighted sagittal view, (B) T2 weighted axial view, cervical-spine level 5.

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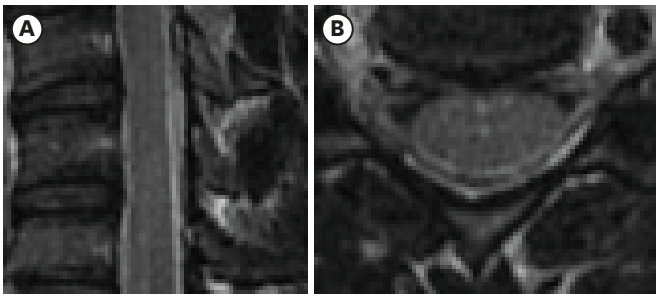

FIGURE 4. Second cervical spontaneous spinal epidural hematoma event 1 month-follow up. A follow-up cervical-spine magnetic resonance imaging on April 11, 2019 showed hematoma regression in Cervical-spine levels 4-6. (A) T2 weighted sagittal view, (B) T2 weighted axial view, cervical-spine level 5.

incidence of SSEH, recurrence of SSEH is much rare, and there are only 11 cases have been reported (**TABLE 1**). The exact mechanism of SSEH has not been identified, and it is still unclear whether the origin of bleeding in acute SSEH is arterial or venous. Some studies postulated that the source of bleeding is the 'free' anastomotic arteries in the epidural space that connect with radicular arteries.[12] Others theorized that SSEH occurs due to local pooling within valve-less, thin-walled epidural veins and brief increases in intravenous pressure caused by intra-thoracic and intra-abdominal pressure elevations, leading to epidural vein rupture.[12] Additional studies in the literature have identified hemorrhages originating in angioma, engorged epidural vessels, venous plexus, or the posterior internal vertebral venous plexus as major causes of SSEH.[14] However, intrathecal pressure is higher than venous pressure, and the sudden onset of symptoms sometimes observed in SSEH is likely not explained by venous theory. Consistently, some studies have identified the cause of SSEH to be of arterial origin.[8,18] Taken altogether, this evidence suggests that there are both venous and arterial origins of SSEH, depending on the case.

Most patients with SSEH present with severe back or neck pain, often with a radicular component. When treatment is delayed, neurologic deficits such as hemiparesis, hemiplegia, quadriparesis, or quadriplegia can be observed. The emergence of neurological symptoms should thus be considered a surgical emergency.[6] Literature suggests that the surgery should be performed within 12–36 hours, depending on the study.[8,12] In addition to surgery, there are some cases of CSSEH that can be resolved with conservative management. A review of medical literature published on PubMed, Embase and Web of Science in January 2018, revealed a total of 17 spontaneously resolved SSEH cases, with radiological imaging proving complete disappearance of the epidural hematoma without surgical intervention.[20] As such, there is still a lack of consensus on whether surgical intervention is necessary to treat SSEH.[18] Interestingly, our patient suffered from CSSEH twice, both of which improved with conservative treatment alone. The patient suffered a sudden-onset paraparesis of motor grade 4 in the first event, and left hemiparesis of motor grade 4 in the second event. In the history taking and evaluation of the patient, he had no remarkable trauma or medical history including surgery, and we could rule out coagulopathy with laboratory findings, and tumor, infection, or vascular malformation with contrast-enhanced MRI.

Zhang et al.[20] reported that for patients with only slight neurologic symptoms, or those showing early and sustained neurologic improvement, non-surgical therapy with close observation is a viable alternative. Considering all reported cases in **TABLE 1**, the American Spinal Injury Association Impairment Scale (AIS) grade D–E was commonly observed

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when hematoma was present in the dorsolateral spine, whereas AIS grade A, indicating a more severe impairment, is more common for hematoma in the ventral spine.[20] This may be because the posterior venous plexus theory of SSEH is unlikely to explain the anterior hematoma.[4] Thus, the ventral hematoma may be arterial in origin, resulting in increased impairment due to the sudden occurrence of symptoms. In such cases, surgery should be considered. In contrast, dorsolateral hematoma is typically accompanied by a less severe impairment and may be responsive to conservative treatment alone. Furthermore, data suggest that conservative treatment may be indicated for patients that showed a tendency towards improved symptoms within 24 hours.

It is important to note that conservative treatment does not always cure SSEH. Morimoto et al.[14] reported 12 cases of recurrence after SSEH onset, and recurrence after conservative treatment was more common. While our patient showed improved neurological symptoms with conservative treatment after both occurrences of SSEH, the possibility of future recurrence and the potential necessity for surgery remains. Nevertheless, if symptoms can be improved without surgery, medical staff and patients will often choose this option to avoid more extensive and invasive surgical approaches.

CONCLUSION

Although surgical intervention is a common course of treatment for SSEH, evidence from our case study and other literature suggest that conservative treatment should be considered when the following three conditions are satisfied: 1) mild neurological symptoms of AIS grade D–E, 2) dorsolateral mass without extensive spinal cord compression and suspected origin in the venous plexus, and 3) spontaneous neurological recovery within 24 hours. If the patient is a candidate to undergo conservative treatment, close observation is necessary to ensure that exacerbation of neurologic deficits do not occur during the recovery period, which would then require prompt surgical intervention.

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