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ABOUT COVER

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Spinal epidural hematoma after spinal manipulation therapy: Report of three cases and a literature review

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Abstract

BACKGROUND

Spinal manipulation therapy (SMT) has been widely used worldwide to treat musculoskeletal diseases, but it can cause serious adverse events. Spinal epidural hematoma (SEH) caused by SMT is a rare emergency that can cause neurological dysfunction. We herein report three cases of SEH after SMT.

CASE SUMMARY

The first case was a 30-year-old woman who experienced neck pain and numbness in both upper limbs immediately after SMT. Her symptoms persisted after 3 d of conservative treatment, and she was admitted to our hospital. Magnetic resonance imaging (MRI) demonstrated an SEH, extending from C6 to C7. The second case was a 55-year-old man with sudden back pain 1 d after SMT, numbness in both lower limbs, an inability to stand or walk, and difficulty urinating. MRI revealed an SEH, extending from T1 to T3. The third case was a 28-year-old man who suddenly developed symptoms of numbness in both lower limbs 4 h after SMT. He was unable to stand or walk and experienced mild back pain. MRI revealed an SEH, extending from T1 to T2. All three patients underwent surgery after failed conservative treatment. The three cases recovered to ASIA grade E on day 5, 1 wk, and day 10 after surgery, respectively. All patients returned to normal after 3 mo of follow-up.

CONCLUSION

SEH caused by SMT is very rare, and the condition of each patient should be evaluated in full detail before operation. SEH should be diagnosed immediately and actively treated by surgery.

Key Words: Spinal epidural hematoma; Spinal manipulation therapy; Spinal cord injury; Magnetic resonance imaging; Surgery; Case report

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Core Tip: Spinal manipulation therapy (SMT) has been widely used worldwide to treat musculoskeletal diseases, but it can cause serious adverse events. Spinal epidural hematoma (SEH) caused by SMT is a rare emergency that can cause neurological dysfunction. We herein report three cases of SEH after SMT, and review a series of literature. All three patients underwent surgery after failed conservative treatment and returned to normal after 3 mo of follow-up. We focused on early recognition and surgical treatment.

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INTRODUCTION

Spinal manipulation therapy (SMT) has been widely used to treat musculoskeletal diseases. The most common indications are neck and back pain. SMT achieves better clinical results than acupuncture or medication[1-3], and complications are relatively rare. However, spinal epidural hematoma (SEH) is a serious complication after SMT, which can cause spinal neurological dysfunction and should therefore be treated. Here, we report three cases of SEH caused by SMT. One patient had a family history of hemangioma, whereas they all had no history of any coagulopathy disease or anticoagulant medication. Platelets and coagulation were normal at the time of admission, and all patients underwent surgery after failed conservative treatment. All patients returned to normal after 3-mo follow-up.

CASE PRESENTATION

Chief complaints

Case 1: A 30-year-old woman was admitted for neck pain and numbness in both upper limbs for 3 d.

Case 2: A 55-year-old man was admitted to the emergency room due to back pain and numbness and weakness of both lower limbs for 1 d.

Case 3: A 28-year-old man suddenly developed numbness and weakness in both lower limbs 4 h after SMT.

History of present illness

Case 1: The patient developed symptoms immediately after SMT 3 d before admission. She took analgesic medications but her symptoms did not completely resolve.

Case 2: The patient experienced sudden back pain after SMT the day before admission, accompanied by numbness and weakness in both lower limbs, an inability to stand or walk, and difficulty urinating. Dehydration medication was given in the community hospital, and the symptoms were not significantly relieved, so he was transferred to our hospital.

Case 3: The patient suddenly developed numbness and weakness in both lower limbs 4 h after SMT. He was unable to stand or walk and had mild back pain, so he was rushed to our emergency room.

History of past illness

None of these three patients had obvious diseases in the past.

Personal and family history

Case three had a family history of hemangioma, and he also had a hemangioma about 1 cm in size, located on the T11. But they all had no history of any coagulopathy disease or anticoagulant medication.

Physical examination

Case 1: Physical examination revealed reduced upper limb sensation and tendon reflexes, normal muscle strength, and grade D spinal cord injury.

Case 2: The sensations, tendon reflexes, and cremaster reflexes of the lower extremities were reduced on both sides. The patient's muscle strength was grade IV, and his spinal cord injury was grade D.

Case 3: The skin sensation below the patient's bilateral nipples was diminished, and muscle strength in the right lower extremity was grade III. The tendon reflex was weakened, the cremaster reflex was not elicited, the anal reflex was weakened, and the spinal cord injury was grade C.

Laboratory examinations

Platelets and coagulation were normal at the time of admission.

Imaging examinations

Case 1: Magnetic resonance imaging (MRI) after admission demonstrated an abnormal shadow in the cervical spinal canal, extending from C6 to C7, and compression of the spinal cord. A T1-weighted image showed high signal intensity, and a T2-weighted image showed low signal intensity that increased on enhanced MRI (Figure 1A-D).

Case 2: Emergency MRI revealed a thoracic spinal canal epidural mass occupying T1 to T3, with spinal cord compression (Figure 2A-C).

Case 3: Emergency MRI revealed a mass lesion in the thoracic spinal canal, extending from T1 to T2, with spinal cord compression (Figure 3A-C), and enhanced MRI indicated signal enhancement (Figure 3D).

FINAL DIAGNOSIS

All three patients were diagnosed with SEH. Blood clots were found during the operation in case 1 and case 2, and the postoperative pathology confirmed a hematoma. In case three, a vein ruptured during the operation, causing massive bleeding.

TREATMENT

Case 1: A laminectomy with internal fixation was performed immediately after admission and the hematoma was removed (Figure 1E). Blood clots were found during the operation, but no bleeding vessels were observed. The postoperative pathology confirmed a hematoma (Figure 1F).

Case 2: After admission, the patient refused surgery and requested conservative treatment, but his symptoms did not resolve. A review of the MRI scan obtained 72 h later revealed that the mass was still compressing the spinal cord (Figure 2D-F). Laminectomy and internal fixation were performed and the hematoma was removed (Figure 2G and H). Blood clots were found during the operation, but no bleeding vessels were observed. The pathological examination suggested a hematoma (Figure 2I).

Case 3: We recommended surgery after admission, but the patient wanted conservative treatment. A 66-h follow-up MRI scan showed a decrease in the mass (Figure 3E), so conservative treatment was continued. The patient suddenly developed paraplegia and complete loss of sensation in both legs after 72 h. A review of the MRI scan indicated fresh bleeding (Figure 3F-H), so an emergency laminectomy was performed and the hematoma was removed. A vein ruptured during the operation, causing massive bleeding; we used hemostatic material to tamp it. Due to the active bleeding during surgery, a specimen could not be obtained for pathological

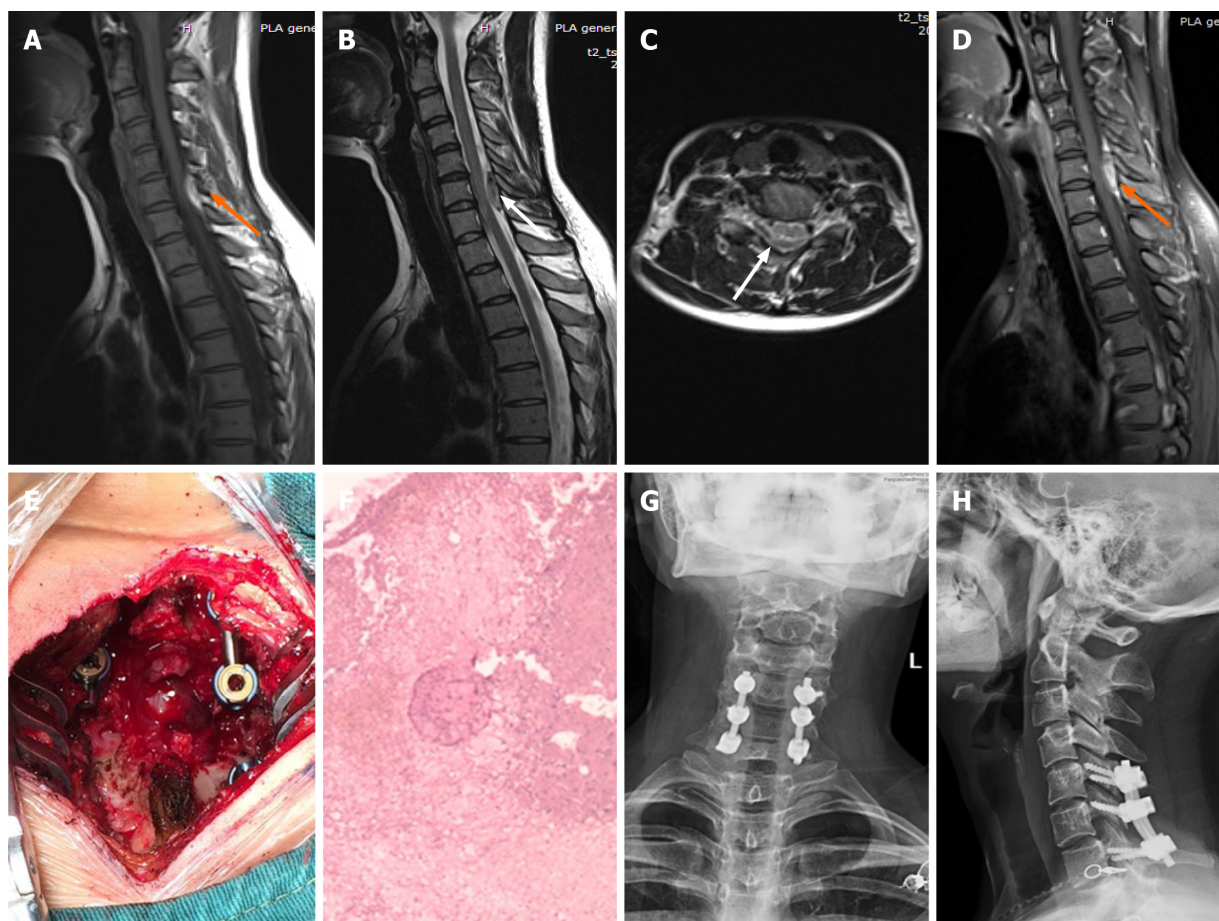


Figure 1 Imaging examinations of case 1. A: T1-weighted preoperative magnetic resonance imaging (MRI) image shows high signal intensity (orange arrow); B and C: Preoperative T2-weighted image shows low signal intensity, and an axial T2-weighted image demonstrates that the hematoma occurred in the posterior region (white arrow); D: Preoperative enhanced MRI suggests an enhanced hematoma signal (orange arrow); E: Intraoperative photograph shows that spinal cord compression has recovered; F: Postoperative pathology suggested a hematoma; G and H: X-ray at the 3-mo follow-up indicated intact internal fixation.

examination.

OUTCOME AND FOLLOW-UP

Case 1: The patient was discharged on postoperative day 5, and her neurological function had fully recovered to grade E. She returned to normal life after the 3-mo follow-up, and the internal fixation position was good (Figure 1G and H).

Case 2: The patient's urine function was fully restored 4 d after surgery. His neurological function recovered to grade E 1 wk after surgery, and he returned to normal life after the 3-mo follow-up.

Case 3: The patient's muscle strength returned to grade V on day 3 after surgery, but he still felt numbness in his feet and had difficulty urinating. The urinary function returned to normal on day 6. His neurological function was grade E on day 10 after surgery. He returned to normal life after 3-mo follow-up, and the hematoma completely disappeared (Figure 3I).

DISCUSSION

SMT is a manual technique for which there is currently no accepted definition, and it is usually performed with a high-speed, low-amplitude thrust that can quickly adjust the joint, often accompanied by a popping sound[4]. Moreover, due to its relatively low invasiveness, many medical and non-medical practitioners use SMT, such as

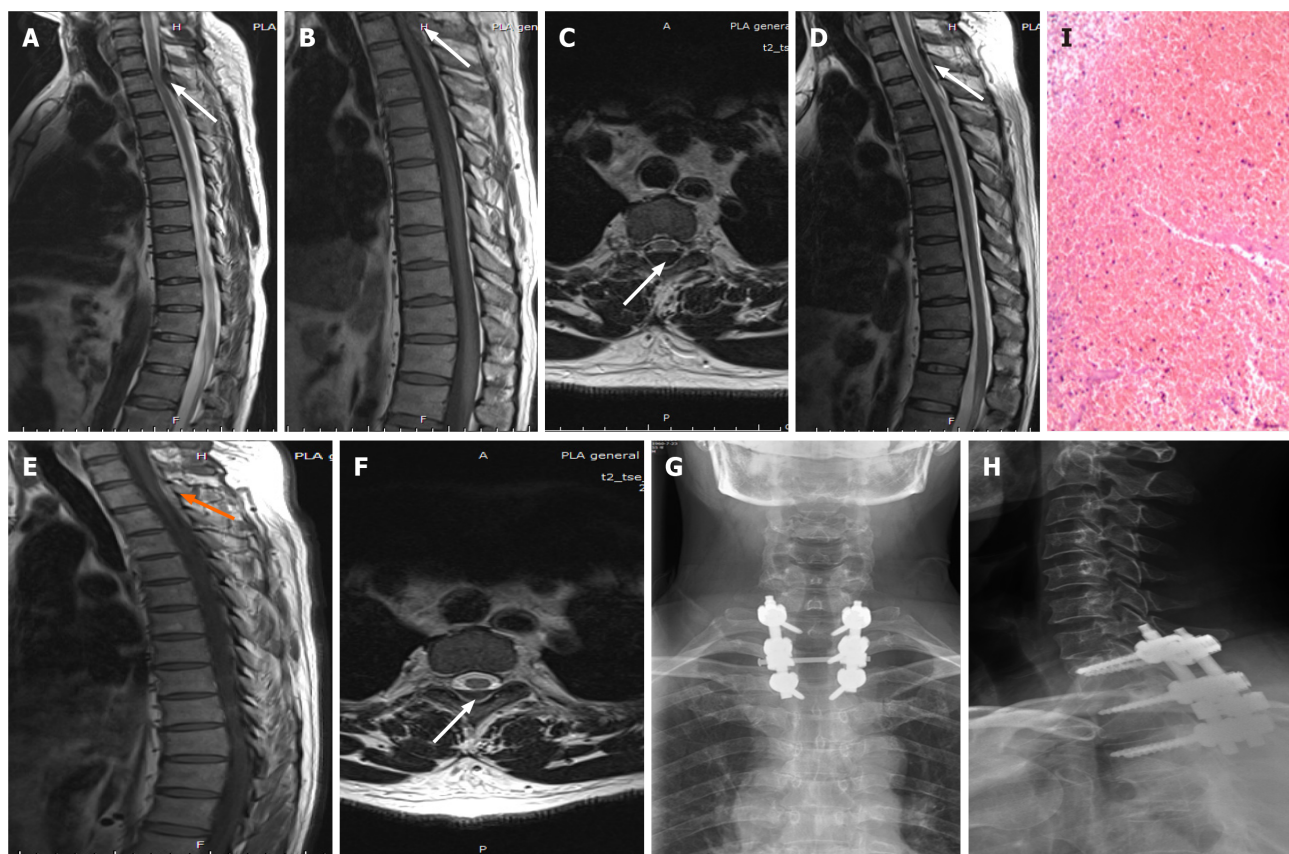


Figure 2 Imaging examinations of case 2. A-C: First emergency magnetic resonance imaging (MRI) images. T1- and T2-weighted images of the thoracic spine show low-intensity spinal epidural hematoma at T1–3; an axial T2-weighted image demonstrates that the hematoma was in the posterior region (white arrow); D-F: The hematoma still existed but was slightly reduced after conservative treatment compared with admission (arrow); G and H: Postoperative review showing good internal fixation; I: Postoperative pathology suggested a hematoma.

chiropractors, osteopaths, and physical therapists[1]. Vertebral artery dissection, spinal cord injury, cauda equina syndrome, cervical subluxation, and cerebrovascular accident can occur even in the hands of experienced therapists, and may cause death in rare cases[5,6]. Patients with chronic coagulopathy deficits, inflammatory spondylosis, osteoporosis, or aortic aneurysm or dissection, and those on long-term anticoagulant therapy are more likely to experience these adverse events[7].

Although many serious adverse events after SMT have been reported, the exact incidence has not been reported. Studies have reported serious adverse event rates of 1 in 20000 and 1 in 250000000 operations. Others reported rates of 1 in 400000 and 1 in 2 million operations[5,8]. Most reports assert that SEH is caused by SMT. We review 18 literature reports (Table 1). Three patients had a history of anticoagulant therapy, two had osteoporosis or spinal degeneration[9–11], and three had a history of spinal degeneration[6,12,13]. There was also one case with an arteriovenous malformation [14]. Of our three cases, one had a family history of hemangioma, whereas they all had no history of coagulation disease or use of anticoagulant medication.

SEH is a rare emergency in which blood accumulates in the spinal epidural space; it begins with back and nerve root pain, and progresses to mild paralysis and urinary retention. In severe cases, SEH may develop into paraplegia or quadriplegia within minutes, hours, or days[15]. Common causes include trauma, lumbar puncture, drug-induced coagulopathy, and genetic coagulation disorders. A few idiopathic cases without a clear etiology have also been reported[16], in addition to clinical cases after minor trauma, such as SMT (Table 1). SEH has been characterized as “idiopathic, spontaneous, and secondary” [16,17]. Most patients develop limb numbness and weakness, severe back pain, and quadriplegia or dyspepsia soon after SMT (up to a few hours later; Table 1), but there have also been reports of delayed symptoms (after 1–3 d[11,18]). All of our cases developed symptoms immediately after SMT, but one case was only detected by examination 3 d later.

The causal relationship between SMT and SEH is often uncertain. Bleeding can occur from a vein or artery. Venous rupture is generally considered to be related to the intravertebral venous plexus[19]. The epidural space of the spinal cord is located in the

Table 1 Summary of cases of spinal epidural hematoma secondary to spinal manipulation therapy

Ref.	Age (yr), sex	Symptoms	Interval to symptom onset	Level	Location of SEH	Treatment	Outcome
Zupruk and Mehta[12], 1989	86, Male	Brown-Sequard Syndrome	24 h	C2-7	Posterolateral	Surgery	Recovery
Segal <i>et al</i> [28], 1996	33, Female	Left upper limb paresis	15 min	C4-6	Posterolateral	Surgery	Recovery
Ruelle <i>et al</i> [25], 1999	64, Female	Paraparesis	2 h	T9-11	Not reported	Surgery	Recovery
Tseng <i>et al</i> [13], 2002	67, Female	Brown-Sequard syndrome	Immediately	C3-5	Posterolateral	Surgery	Recovery
Saxler and Barden[31], 2004	27, Female	Headache	10 min	C1-S1	Not reported	Conservative	Recovery
Whedon <i>et al</i> [9], 2006	79, Male	Lower extremity paralysis	Immediately	C4-5	Posterolateral	Surgery	Recovery
Gouveia <i>et al</i> [26], 2007	34, Male	Tetraplegia	Few hours	C3-6	Not reported	Surgery	Recovery
Domenicucci <i>et al</i> [16], 2017	52, Female	Brown-Sequard Syndrome	Immediately	C3-T1	Posterolateral	Surgery	Recovery
Solheim <i>et al</i> [10], 2007	77, Male	Right lower extremity paresis	Immediately	L3-4	Posterior	Surgery	Partial recovery
Heiner[32], 2009	38, Female	Neck and arms pain numbness	4 h	C1-4	Posterolateral	Conservative	Recovery
Lidder <i>et al</i> [21], 2010	64, Male	Hemiparesis	Immediately	C4-6	Not reported	Surgery	Partial recovery
Lee <i>et al</i> [27], 2011	38, Female	Lower extremity paralysis	Immediately	T1-7	Posterolateral	Surgery	Recovery
Huang <i>et al</i> [14], 2015	40, Male	Hemiparesis	Immediately	C2-T2	Posterolateral	Surgery	Recovery
Ling <i>et al</i> [6], 2017	33, Male	Tetraplegia	Immediately	C4-7	Posterolateral	Surgery	Die
Fattahi <i>et al</i> [30], 2017	44, Female	Tetraplegia	not reported	C1-4	Anterior	Conservative	Recovery
Ryu <i>et al</i> [29], 2018	38, Male	Paraparesis	Immediately	C6-T1	Anterior	Conservative	Recovery
Vanichkulbodee <i>et al</i> [18], 2019	20, Male	Paraparesis	3 d	C6-T2	Posterolateral	Surgery	not reported
Cooper <i>et al</i> [11], 2019	63, Male	Tetraplegia	1 d	C6-7	Posterior	Surgery	Partial recovery
Present case one	30, Female	Neck and arms pain numbness	Immediately	C6-7	Posterolateral	Surgery	Recovery
Present case two	55, Male	Lower extremity paralysis	1 d	T1-3	Posterior	Surgery	Recovery
Present case three	28, Male	Paraparesis	4 h	T1-2	Posterolateral	Surgery	Recovery

SEH: Spinal epidural hematoma.

spinal canal. It contains loose areolar tissue and an extensive internal venous plexus that receives projections from the vertebral bodies and spinal cord. This internal venous plexus communicates freely with the external venous plexus, which together form Batson's veins. This low-pressure system without valves is particularly susceptible to sudden changes in pressure, such as when lifting, straining, coughing, or vomiting[15]. When performing SMT, a low-amplitude thrust will produce sufficient pressure in the spine for rupture of fragile veins, which in turn will lead to clinical symptoms of compression of the spinal cord. However, it has been posited that the hematoma originates from an artery[20]. Lidder *et al*[21] observed ruptured veins during the operation. We detected venous bleeding during surgery in a patient with a family history of hemangioma, but could not find the bleeding source. Hemostatic materials were used to stop the bleeding. Although the cause of bleeding was not clarified, timely diagnosis and early treatment are very important for SEH.

MRI has advantages for diagnosis of an SEH. The hematoma is usually isointense on T1-weighted images and hyperintense on T2-weighted images within 24 h of the onset of symptoms. The hematoma is often hyperintense on T1- and T2-weighted images after 24 h, while a chronic hematoma becomes hypointense on both T1- and T2-weighted images. The hematoma usually shows significant enhancement[22]. In both

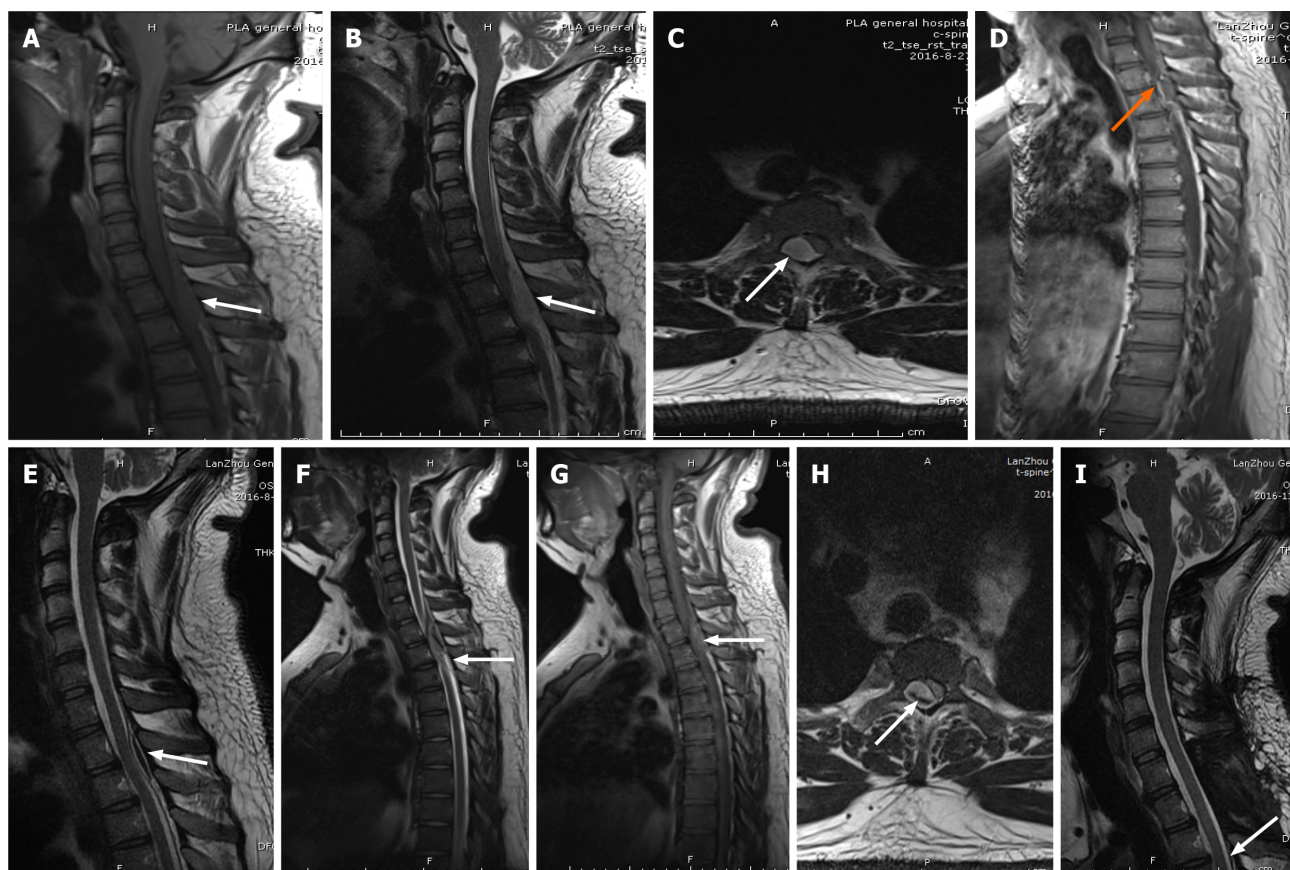


Figure 3 Imaging examinations of case 3. A: First emergency magnetic resonance imaging (MRI) scan. T1-weighted image of the thoracic spine shows isometric-intensity spinal epidural hematoma at T1–2 (white arrow); B and C: First emergency MRI scans. T2-weighted image shows high signal intensity; axial T2-weighted image shows that the hematoma was in the posterolateral region (white arrow); D: Preoperative MRI suggests an enhanced hematoma signal (orange arrow); E: The hematoma decreased in size after conservative treatment (white arrow); F–H: After 72 h of conservative treatment, MRI showed that the hematoma worsened again (white arrow); I: The hematoma completely disappeared at the 3-mo follow-up (white arrow).

our first and third cases, MRI showed enhancement.

Most people believe that surgery is still the first choice for treating SEH. Liao *et al* [23] reported that surgery for SEH is safe and effective. When performed within 48 h after the initial attack, complete neurological dysfunction is recommended not to exceed 12 h [23]. Groen and van Alphen [24] reported that surgery was performed within 36 h in patients with complete neurological dysfunction, and within 48 h in those with incomplete neurological dysfunction [24]. The procedure involves decompressive laminectomy and evacuation of the hematoma. Most patients in the literature underwent surgery at the time of symptom onset or exacerbation, and postoperative recovery was good [12,14,25–27]. However, three patients underwent surgery after a delay of several days; their symptoms did not completely resolve [10,21,28], and one patient died from complications [6]. Although the majority of studies reported good recovery after surgical excision of SEH, there have also been reports that conservative treatments can provide good results [29–32]. Fat tissue in the epidural space contains a huge network of capillaries that usually absorbs foreign substances; thus, the SEH should be absorbed over time. However, irreversible nerve damage can occur during absorption [33]. Conservative treatment was preferred in all of our cases, but the nerve injury symptoms did not completely disappear. Two patients were operated after conservative treatment proved ineffective; one patient developed sudden paraplegia during conservative treatment and was immediately operated in the emergency department. The symptoms disappeared after the operation.

Although surgery can achieve good results, clinical follow-up is needed to prevent the occurrence of complications. To avoid SEH after SMT, good planning and individualized assessment are needed, and risk analysis should be performed based on the results of multiple clinical studies [34].

CONCLUSION

In conclusion, before proceeding with SMT, each patient should be evaluated in detail and checked for risk factors. In cases where the physical condition changes rapidly, physicians should be alert to the danger and send the patient to the emergency department for a complete MRI examination. We recommend surgery if neurological symptoms appear.

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