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Case Report

Anticoagulant induced spontaneous spinal epidural hematoma, conservative management or surgical intervention—A dilemma?

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Abstract

Spontaneous spinal epidural hematoma (SSEH) is a rare cause of cord compression. SSEH with neurological deficit is an emergency situation that is commonly considered an indication for emergency surgical decompression. We describe a patient with SSEH who recovered clinically and radiologically with conservative treatment. A 25-year-old hypertensive male presented with acute onset back pain followed by asymmetrical paraparesis. He had sensory level at D9 dermatome with preserved bladder and bowel functions. He was taking anticoagulants for deep venous thrombosis of the left lower limb. Surgery was deferred because of the deranged coagulation profile. He was managed conservatively with correction of coagulopathy. After 3 days, he recovered significantly. Repeat neuroimaging revealed significant resolution of epidural hematoma. The conservative approach can be considered for selected patients who are unsuitable for early surgical intervention, those with stable neurological status [American Spinal Injury Association (ASIA) Scale E], or those in whom early recovery of function has been initiated with ASIA Scale C or D. Neurological status at presentation and suitability for surgical intervention seem to be important determinants of the type of therapeutic intervention.

Keywords: anticoagulant; conservative management; spinal epidural hematoma; spontaneous spinal epidural hematoma

1. Introduction

Spontaneous spinal epidural hematoma (SSEH) is a rare condition with an estimated incidence of 0.1 per 100,000 patients per year.[1] The etiology of SSEH is usually uncertain. The predisposing factors include vascular malformations, neoplasm, anticoagulation therapy, antiplatelet drugs, systemic hypertension, pregnancy, coronary thrombolysis, and very rarely following lumbar puncture.[2–4] In half of the cases, no predisposing factor is identified despite extensive evaluation. SSEH usually presents acutely with complaints such as neck pain or back pain, radiating pain, mild sensorimotor deficits, bladder or bowel symptoms, and even complete paralysis.[5–7] Occasionally, SSEH may present as cauda equina syndrome due to compression of spinal nerve roots.[5] Other disorders such as epidural tumor, epidural abscess, and acute disk herniation may mimic SSEH clinically. The most common presentation is acute onset back pain and radicular symptoms that may mimic disk herniation. Because SSEH may lead to severe neurological deficit and morbidity, prompt diagnosis and treatment are important. The primary treatment modality in SSEH is decompressive surgery, as a delay in intervention may result in a poor clinical outcome. Although the number of reports in the literature is increasing regarding spontaneous resolution of the SSEH, no evidence-based guideline is available on the optimal treatment of the

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SSEH.[8–15] We report a case of anticoagulant-induced SSEH that showed spontaneous rapid resolution of the SSEH on conservative management.

2. Case Report

A 54-year-old hypertensive man presented with sudden onset back pain followed by weakness of both lower limbs for 1 day. Weakness was felt more in the right lower limb compared with the left. He required the support of one person to walk due to weakness. He had a history of pain and swelling of the left lower limb 4 months previously, and was diagnosed to have deep venous thrombosis (DVT) on venous Doppler examination. Subsequently, he was placed on anticoagulant (warfarin, 7.5 mg/day) therapy. He had no history of falls or trauma to the back preceding the lower limb weakness. He denied any drug allergy or addiction. He was alert and fully oriented. The clinical examination revealed asymmetrical weakness of both lower limbs. The motor examination showed Medical Research Council (MRC) Grade 2 power proximally and Grade 3 power distally in the right lower limb. In the left lower limb, he had MRC Grade 4 power proximally as well as distally. He had American Spinal Injury Association (ASIA) impairment scale Grade C (details of the ASIA scale are shown in Table 1).[16] The sensory examination revealed the sensory level at D9 dermatome with sensory loss for pain, touch, and temperature in both lower limbs. Joint position sensations were impaired in both lower limbs. Deep tendon reflexes were suppressed in both lower limbs. The rectal tone was normal. He had no urinary complaints. There was no neurological deficit in both upper limbs.

Magnetic resonance imaging (MRI) revealed epidural hematoma in the dorsal aspect of the spinal canal extending from D3 to D8 levels, causing compression on the cord and displacing it anteriorly with secondary central canal stenosis at D5–D6 to D7–D8 levels (Figure 1). Blood investigations showed a prothrombin time of 77.3 seconds, an international normalized ratio of 5.93, and an activated partial thromboplastin time of 90 seconds. Blood cell counts, erythrocyte sedimentation time, liver function test results, and renal function test results were within normal limits. A venous Doppler of the left lower limb showed chronic DVT with partial recanalization. In view of the sensorimotor deficit with significant cord compression, neurosurgery opinion was sought and urgent decompression was recommended. As he had a deranged coagulation profile because of the anticoagulant, four units (15 mL/kg) of fresh frozen plasma (FFP) was given. Within 6 hours of FFP infusion, his weakness started to improve with one grade. Conservative treatment rather than neurosurgical intervention was considered in view of the improvement in neurological status. On Day 3 of hospital stay, he started to walk independently. Sensory deficit resolved completely. Repeat neurological examination revealed MRC Grade 5 power proximally and +4/5 distally in both lower limbs. An MRI of the spine was repeated after 5 days and revealed significant resolution of the epidural hematoma (Figure 2). After 7 days, he was again started on anticoagulant for DVT with monitoring of the coagulation profile. At 3 months of follow-up, he was independent without any neurological deficit.

Table 1
American Spinal Injury Association (ASIA) impairment scale.

ASIA scale	Degree of neurological impairment
A	Complete: no motor or sensory function preserved
B	Incomplete: sensory but no motor function preserved below the neurologic level
C	Incomplete motor function (>50%) of the key muscles below the neurologic level, motor grade < III
D	Incomplete motor function (>50%) of the key muscles below the neurologic level, motor grade ≥ III
E	Normal

3. Discussion

The underlying pathogenesis of SSEH is not clear.[2,3,17] The most important factor identified as contributing to SSEH is a hemorrhagic diathesis from either medications or disease states.[18,19] Only on rare occasions, SSEH is associated with an identifiable underlying vascular lesion. Various medications have been reported to be associated with SSEH. These include antiplatelet, anticoagulant, or thrombolytic medications, such as aspirin, warfarin, heparin, tissue plasminogen activator, and streptokinase.[20] Anticoagulants are mainly used in secondary prophylaxis of DVT, cerebral embolism secondary to lone atrial fibrillation or valvular heart disease, and cortical sinus thrombosis. Anticoagulation and/or thrombolytic therapy is also used in treatment of acute myocardial infarction.[20] In our case, the patient was taking warfarin for secondary prophylaxis of DVT. Anticoagulants, including warfarin, are used in 25–70% of patients with SSEH and are an important risk factor.[21,22] Coagulation variables are within the therapeutic range in many of the reported cases of SSEH.[21] In such cases, epidural bleeding may be initiated because of other factors. In addition to use of anticoagulant agents, the relationships with hypertension, some structural extradural anomalies, the rupture of fragile epidural veins by an adjacent herniated disk, trivial trauma, and straining are suggested.[21,23–25] Omori et al[26] pointed out the possibility of the involvement of arterial problems and cervical spondylitis.

One hypothesis suggests that elevated intrathoracic and/or intra-abdominal pressure results in rupture of the vertebral venous plexus.[27] The most frequent reported site of SSEH is the cervicothoracic region with the predominant involvement of the dorsal side of the spinal canal.[23,28] The posterior internal vertebral venous plexus in the cervicothoracic region is bigger and more convoluted compared with the anterior one. Beyond this, the posterior venous plexus is uncovered by ligamentous structures. Therefore, this hypothesis seems to be important for the underlying mechanism of SSEH.[28] Another hypothesis postulates arterial rupture as an origin of SSEH as the intrathecal pressure is greater than the vertebral venous plexus.[29] In our case, SSEH was located in the dorsal aspect of

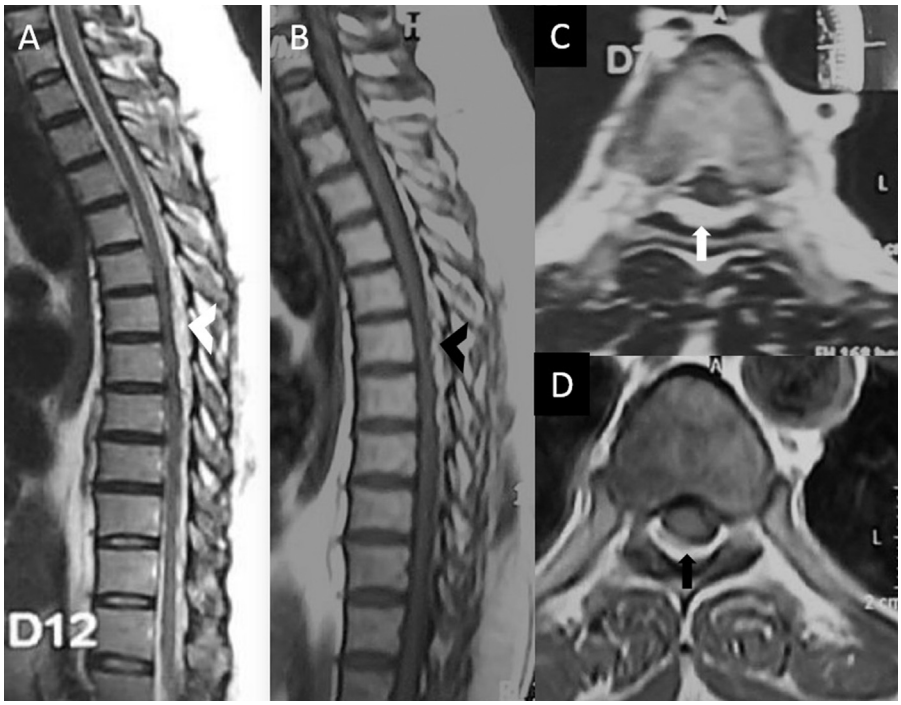

Figure 1. (A) T2-weighted sagittal sequence [4000/98/2 (TR/TE/excitations)], obtained 18 hours after the event, shows the hyperintense epidural lesion (white arrow head) in the dorsal aspect of the thoracic spinal canal extends from D5–D6 to D7–D8 level. The epidural lesion is isointense but slightly heterogeneous compared with CSF. The low signal intensity of the dura separating the lesion and cord is clearly seen; (B) T1-weighted sagittal sequence (600/8/2) shows hyperintense epidural lesion (black arrowhead); (C) axial T2-weighted (4500/96/2) and (D) axial T1-weighted (500/9/2) MRI at the level of D7 show hyperintense lesion (white and black arrow, respectively) posterolaterally in dorsal spinal canal causing compression and displacement of the cord anteriorly. CSF = cerebrospinal fluid; MRI = magnetic resonance imaging; TE = echo time; TR = repetition time.

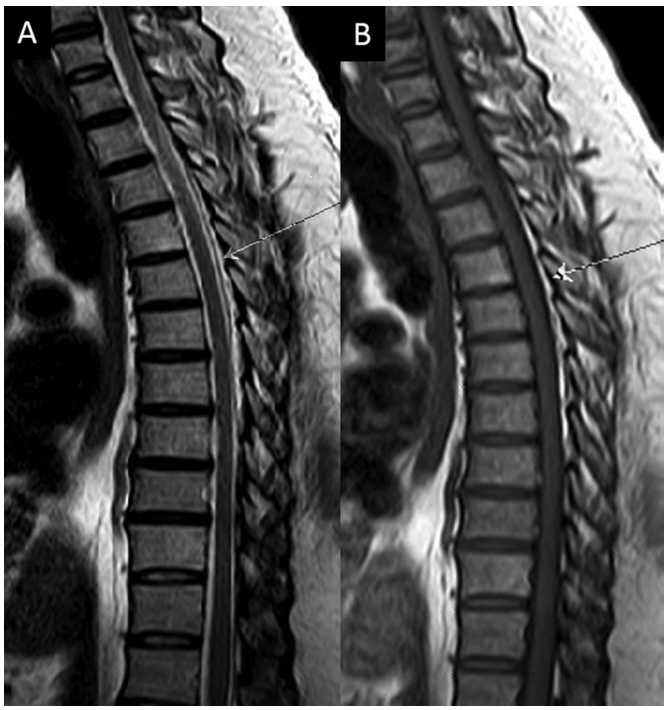

Figure 2. (A) Follow-up midthoracic sagittal T2-weighted (4000/98/2) and (B) T1-weighted (400/15/2) sequences, obtained 5 days after the initial imaging, reveal nearly complete resolution of hyperintense epidural collection extending from D5–D6 to D7–D8 levels (long thin arrows) compared with Figures 1A and 1B.

the thoracic spinal canal. This emphasizes the first theory of origin of SSEH.

MRI is the diagnostic method of choice for SSEH.[5] If MRI is unavailable, computed tomography scans should be obtained. MRI provides information about the location, extent of the hematoma, as well as the degree of cord compression. The differential diagnosis of SSEH includes acute herniated intervertebral disk, epidural tumor or abscess, spinal abscess, spinal cord ischemia, and transverse myelitis. MRI recognition of blood products helps in distinguishing SSEH from other spinal extramedullary lesions. MRI typically shows biconvex hematomas in the epidural space with well-defined borders tapering superiorly and inferiorly. In addition, MRI may rarely demonstrate underlying lesions, such as spinal vascular malformations.

Holtås et al[1] examined the MRI characteristics of SSEH in a series of 13 patients from 1986 to 1995. In this group, five patients had minor trauma and four were receiving anticoagulant therapy. The most common location was the upper thoracic region. T1-weighted images were found to be most useful in following the signal shift from T1 isointensity to T1 hyperintensity as the hematoma evolved from acute to subacute.

Early surgical intervention has been traditionally advocated as a treatment of choice for SSEH.[6,30] The procedure includes decompressive laminectomy and hematoma evacuation. Time of the procedure from the onset of symptoms has been considered the most crucial factor for neurological recovery. Most of the authors advocate the procedure within 36 hours if the neurological deficit is complete and within 48 hours in case of incomplete

neurological deficit.[31,32] The clinical outcome correlates with the severity of neurological deficit preoperatively.[31,33]

Recently, many reports have appeared in the literature regarding the spontaneous resolution of SSEH without surgery. In most of the reports, the authors chose conservative treatment in the case of rapid improvement of neurological deficits, inappropriateness for surgery such as coagulopathy, and the refusal of surgery.[9–14] In a comparative study of surgical versus conservative management of SSEH, one-third ($n = 5$) of 15 patients were managed conservatively.[15] There was no significant difference between the two groups. The final clinical outcome was correlated with the initial neurological status. The study emphasized conservative management in patients whose present neurologic status was ASIA Scale E or in whom early recovery of function has been initiated with ASIA Scale C or D. In a review of published literature, only 10 out of 64 conservatively treated patients had incomplete recovery.[10] It can be explained by milder neurological deficit at presentation in conservatively treated patients in comparison to those who were surgically treated.

Our case had coagulopathy secondary to anticoagulant therapy (warfarin). Initially, surgery was deferred because of the deranged coagulation profile. Within 6 hours of correction of coagulopathy, the patient started to improve. In view of the improvement of Grade 1, the decision to use conservative treatment was taken.

The National Acute Spinal Cord Injury Studies II and III, a Cochrane database of systematic reviews of all randomized clinical trials, and other published reports have recommended the use of high doses of methylprednisolone within 8 hours of acute spinal injury.[34–36] Recently, these studies were revisited and the validity of their results was questioned. The updated guidelines issued in 2013 by the Congress of Neurological Surgeons and the American Association of Neurological Surgeons recommend against the use of steroids early after an acute spinal cord injury.[37] There is no randomized trial on the use of steroid in SSEH. In some of the studies, a low dose of steroid was used in SSEH.[15] In view of the side effects of steroid therapy such as increased risk of infection, gastric hemorrhage, and avascular necrosis, and the lack of evidence of the benefits of steroid therapy, at present steroid therapy cannot be recommended for the treatment of SSEH. Steroid therapy was not used in our case. The improvement noted in our case without steroid therapy questions the utility of steroid therapy in SSEH.

No randomized controlled trial is available on the comparison of conservative versus surgical management of SSEH. The rarity of SSEH cases and the ethical problems encountered in randomly allotting the patients with neurological impairment secondary to SSEH to conservative management are the main hurdles in designing a prospective randomized controlled trial.[15]

In the pediatric population, literature is available on SSEH in hemophilic children. The recent literature endorses the conservative management in SSEH with stable neurological status in hemophilic children.[38–41] High-risk surgical intervention with inappropriate coagulation status should be avoided in such cases unless neurological deficit progress rapidly. No such literature is available in the adult population with coagulopathy.

4. Conclusion

It is important for all clinicians to include SSEH in the differential diagnosis of acute focal neurologic deficit, especially when associated with sudden and unexplained neck or back pain and in the setting of anticoagulant therapy or hemorrhagic diathesis. Multiplanar spinal MRI is optimal for the diagnosis and assessment of the extent of the lesion and degree of spinal cord compression. Early decompressive surgery is the recommended approach in the management of SSEH especially in the presence of rapidly progressive neurological deficit. However, the conservative approach can be adopted in patients who are unsuitable for early surgical intervention, who present with stable neurological status (ASIA Scale E), or in whom early recovery of function has been initiated with ASIA Scale C or D. Neurological status at presentation and suitability for surgical intervention seem to be the important determinants for the type of therapeutic intervention to be used. Further studies are necessary to explore other factors to determine the management approach in patients with SSEH.

Conflicts of interest

All authors have no conflicts of interest to declare.

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