Clinical Case Report

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Nonsurgical management of an extensive spontaneous spinal epidural hematoma causing quadriplegia and respiratory distress in a choledocholithiasis patient

A case report

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Abstract

Rationale: Spontaneous spinal epidural hematoma (SSEH) manifests from blood accumulating in the epidural space, compressing the spinal cord, and leading to acute neurological deficits. The disease's cloudy etiology and rarity contribute to dangerously suboptimal therapeutic principles. These neural deficits can be permanent, even fatal, if the SSEH is not treated in a timely and appropriate manner. Standard therapy is decompressive laminectomy, though nonsurgical management is a viable course of action for patients who meet a criterion that is continuously being refined.

Patient concerns: A 76-year-old woman on warfarin for a past pulmonary embolism presented to the emergency room with jaundice, myalgia, hematuria, neck pain, and an International Normalized Ratio (INR) of 14. Upon admission, she rapidly developed quadriplegia and respiratory distress that necessitated intubation.

Diagnoses: T2-weighted magnetic resonance imaging (MRI) revealed an epidural space-occupying hyperintensity from C2 to S5 consistent with a spinal epidural hematoma. An incidental finding of dilated intrahepatic and common bile ducts prompted an endoscopic retrograde cholangiopancreatography, which demonstrated choledocholithiasis.

Interventions: The patient's INR was normalized with Vitamin K and Beriplex. Upon transfer to the surgical spine team for assessment of a possible intervention, the patient began to demonstrate recovery of neural functions. The ensuing sustained motor improvement motivated the team's preference for close neurologic monitoring and continued medical therapy over surgery. Thirteen hours after the onset of her symptoms, the patient was extubated. A sphincterotomy was later performed, removing 81 common bile duct stones.

Outcomes: MRI demonstrated complete resorption of the SSEH and the patient maintained full neurological function at final follow-up.

Lessons: Nonsurgical management of SSEH should be considered in the context of early and sustained recovery. Severe initial neural deficit does not necessitate surgical decompression. Choledocholithiasis and subsequent Vitamin K deficiency, particularly when coupled with anticoagulant use, can increase INR and is a novel proposed risk factor for SSEH. Furthermore, coagulopathies should be medically corrected before surgical intervention within a given timeframe, as spontaneous recovery may manifest. This should be favored over surgery in patients demonstrating early and sustained recovery, as nonsurgical management is 25% more effective in achieving full recovery.

Abbreviations: ASIA = American Spinal Injury Association, ERCP = endoscopic retrograde cholangiopancreatography, INR = international normalized ratio, MRC = Medical Research Council, MRI = magnetic resonance imaging, SSEH = spontaneous spinal epidural hematoma.

Keywords: anticoagulation, choledocholithiasis, nonsurgical management, paralysis, respiratory distress, reversible coagulopathy, spontaneous spinal epidural hematoma

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The authors report no conflicts of interest.

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1. Introduction

Spontaneous spinal epidural hematoma (SSEH) is a rare pathology characterized by blood accumulating in the epidural space and compressing the spinal cord,[1]. The resulting sensorimotor deficits can range from back pain to quadriplegia.[2] The term "spontaneous" refers to the atraumatic etiology and contributing multifactorial factors, such as oral anticoagulant use.[3,4] Investigation of choice is magnetic resonance imaging (MRI).[5] The degree of pre-therapy neural deficit is a major prognostic factor.[6] Suboptimal therapeutic principles contribute to SSEH's 5.7% mortality[7] and a morbidity 10 times as high.[8] Standard therapy is urgent decompressive laminectomy, though nonsurgical management is a viable course of action often overlooked in current literature.[9] We present a unique case of severe SSEH secondary to choledocholithiasis that was treated nonsurgically with full recovery.

2. Case report

A 76-year-old female taking warfarin due to a past pulmonary embolism presented to the emergency department jaundiced with a 3-day history of diffuse atraumatic myalgia and macroscopic hematuria. Laboratory evaluation revealed an international normalized ratio (INR) of 14.04. She was consequently administered Vitamin K. Initial neurovascular examination was normal, though within 10 minutes, the patient deteriorated with an acute onset of flaccid quadriplegia, graded as an American Spinal Injury Association (ASIA) score of A. This was accompanied by severe neck pain and respiratory distress with an O_2 saturation of 73%. She was intubated, administered Beriplex, and continued to receive Vitamin K. Within 2 hours, her INR normalized to 1.60 with motor functioning limited to flickers in her toes and right fingers. An MRI performed 4 hours after symptom onset demonstrated an SSEH from C2 to S5, causing significant posterior spinal cord displacement, central canal stenosis, spinal and cauda equina nerve root compressions, seen in Fig. 1. The patient was transferred to the orthopedic spine team for assessment of a possible intervention, where she began to demonstrate recovery of neural functions. By 11 hours after the onset of her symptoms, the patient had improved to an ASIA score of D, with a Medical Research Council (MRC) grading of 4+ in her upper limbs and 4 in her lower limbs. The ensuing sustained motor improvement motivated the team's preference for close neurologic monitoring and continued medical therapy over surgery. She continued to improve and was extubated 13 hours after symptom onset. By 20 hours, the patient had regained full motor and sensory functions, returning to her initial neurological baseline at an ASIA score of E. Dilated intrahepatic and common bile ducts were incidentally detected on MRI, prompting an endoscopic retrograde cholangiopancreatography (ERCP). The ERCP demonstrated obstructions in the biliary tree consistent with choledocholithiasis, as seen in Fig. 2. Subsequent sphincterotomy removed 81 common bile duct stones. MRI performed a month after symptom onset demonstrated complete resorption of the SSEH without spinal surgical intervention as seen in Fig. 3. The patient maintained full neurological function at final follow-up 3 months after the nonsurgical resolution of her SSEH. The patient's informed consent for this report was obtained and further approved by McGill University Health Centre's Ethics Review Board.

3. Discussion

When confronted with SSEH's potentially rapid and debilitating onset of symptoms, urgent surgical decompression may seem like a given. However, we present a case of extensive hematoma causing severe neurological compromise that responded to nonsurgical management, demonstrating its efficacy in a wider array of SSEH presentations than previously thought. Conservative management is a recent recommendation for SSEH patients presenting with minimal neural deficits or when spontaneous recovery is observed.[9] Nonsurgical management is 25% more effective than surgery in achieving complete recovery, regardless of neural deficit upon presentation.[8] We recommend that patients presenting with severe neural deficits should not be excluded from nonsurgical management, though decompressive laminectomy should remain readily available.[10] The decision to manage our patient nonsurgically was influenced by her medically correctable INR, progressive recovery, and the extensive size of hematoma. The inherent risks of nerve damage,

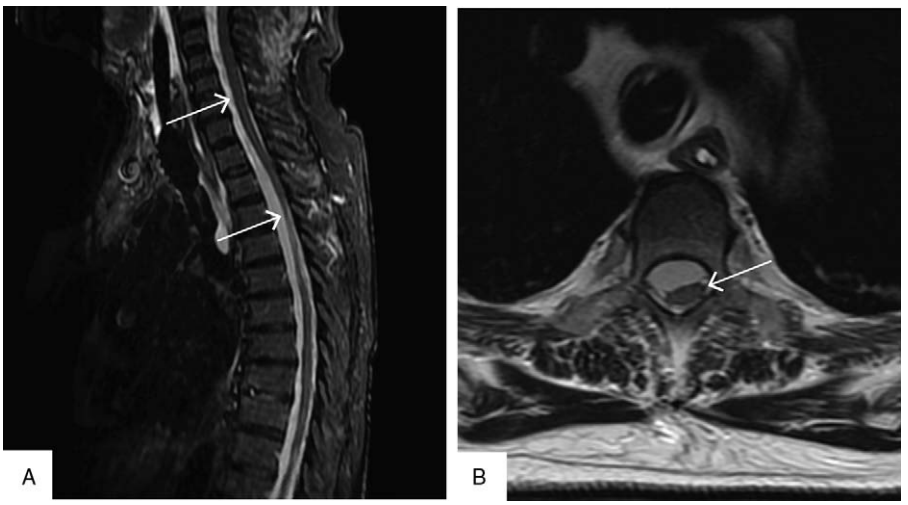

Figure 1. T2-weighted MRI images demonstrating an epidural hematoma. Arrows indicate significant spinal cord displacement. (A) Sagittal view of SSEH in cervicothoracic spine with C2 as the first visible vertebrae. (B) Axial view of SSEH in the thoracic spine.

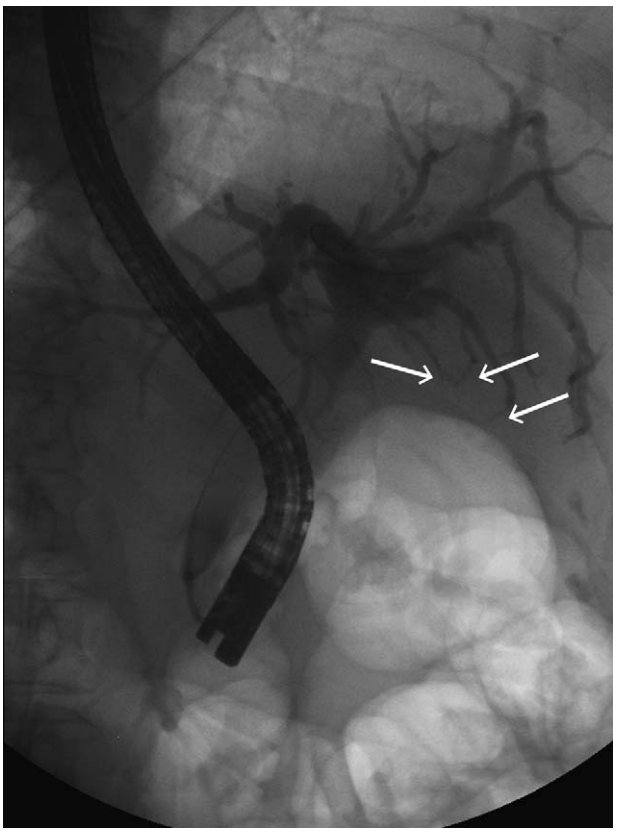

Figure 2. Endoscopic retrograde cholangiopancreatography image demonstrating choledocholithiasis. Arrows indicate opacifications indicative of obstructions, consistent with stones in the biliary tree.

leaks, and infections associated with multilevel laminectomies required to decompress such an extensive SSEH further dissuaded surgical intervention, given the sustained motor improvement observed manifesting nonsurgically.

Our patient highlights both a known and a potentially novel spontaneous hematoma etiology. Oral anticoagulant use is a well-described risk factor for SSEH.[11] Our patient's warfarin regime contributed to her supratherapeutic INR of 14.04, predisposing her to hemorrhage, though it is unlikely to be solely responsible. Choledocholithiasis and the resulting obstructive jaundice have been shown to majorly contribute to Vitamin K deficiency.[12] This well-established pathologic process would be reflected by a significant increase INR,[13] and, as our patient demonstrates, consequently increase the risk for SSEH. Spontaneous recovery from SSEH is thought to be due to the gradual spread of hematoma throughout the epidural space and along exiting nerve roots in adjacent, highly vascularized fatty areolar tissue,[14] thereby decompressing the spinal cord and decreasing neural deficits.[15] The patient's hematoma resolved only after her INR normalized to 1.60. Perhaps the coagulopathy's reversibility contributed to the observed effectiveness of nonsurgical management. This is the first known case of SSEH secondary to both anticoagulation therapy and choledocholithiasis, and we propose that the latter be considered a novel potential risk factor for SSEH in patients on warfarin.

In conclusion, SSEH is a rare and debilitating pathology that can resolve with nonsurgical management regardless of the severity of symptoms, given certain patient characteristics exist. Coagulopathies should be medically corrected before surgical intervention within a given timeframe, as spontaneous recovery may manifest. This timeframe would depend on the initial severity of neural deficit caused by the SSEH, the presence or absence of spontaneous recovery beginning to manifest, and the amount of time required for the coagulopathy to correct. The atraumatic etiology, absence of surgical initiating factors, widespread affected anatomical territory, severe degree of initial neural deficit, and complete recovery all make this SSEH case an ideal model to study the reversibility of acute spinal cord compression symptoms. By continuing the collection of conservatively managed cases, we can identify the critical timeframe until irreversible damage occurs,[16] indicate the likelihood of spontaneous recovery, and create a better risk–benefit metric for when it is best to surgically intervene and, as was the case with our patient, when it is best to not.

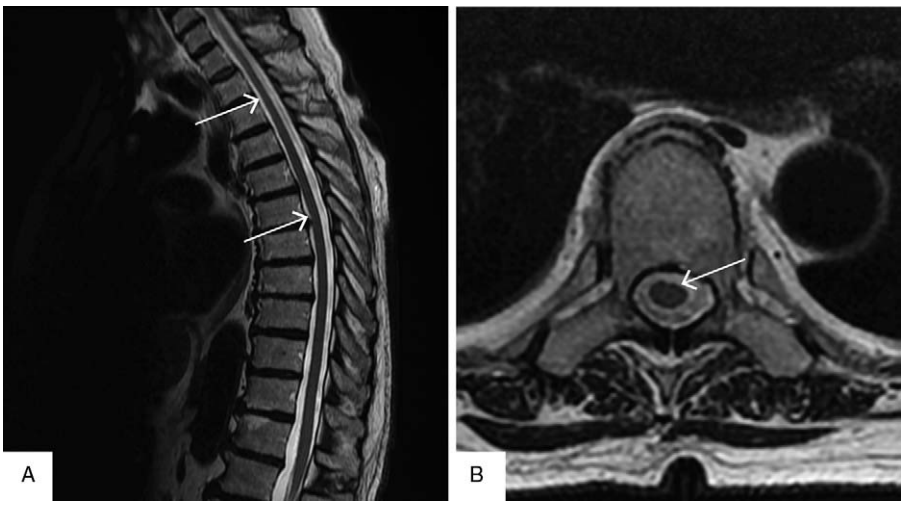

Figure 3. T2-weighted MRI images demonstrating the spontaneous and complete hematoma resorption without surgical intervention. Arrows indicate the return to normal spinal anatomy. (A) Sagittal view of the thoracic spine with T2 as the first visible vertebrae. (B) Axial view in the thoracic spine.

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