A spontaneous cervical spinal epidural hematoma in a male patient receiving treatment for acute coronary syndrome: A case report

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ABSTRACT

Although spontaneous spinal epidural hematoma is a rare entity with an unknown origin, it may occur secondary to the use of anticoagulant and/or antiplatelet agents, which are particularly used for the treatment of cardiovascular and cerebrovascular diseases. Since it occurs rarely and its initial symptoms are usually non-specific, early and accurate diagnosis can be challenging which affects survival rate and the quality of life. Herein, we present a 65-year-old male case who developed acute severe neck pain and headache on the third day of acute coronary syndrome treatment, followed by neurological deficits in bilateral upper and lower extremities.

Keywords: Anticoagulants, laminectomy, neck pain.

Spontaneous spinal epidural hematoma (SSEH) is a very rare disease warranting early diagnosis and treatment. Although its prevalence is predicted to be 1 in 100,000, it has a high risk of morbidity and mortality. Despite its uncertain pathogenesis, predisposing factors have been reported to be coagulation disorders, vascular malformations, hypertension, cancer, pregnancy and, most commonly, anticoagulant and antiplatelet treatments. However, no predisposing factor can be detected in 40% of the patients reported in the literature. The most common clinical presentation is the emergence of nerve root and spinal cord compression symptoms accompanying or following sudden-onset spinal pain. On the other hand, initial symptoms can appear in a wide range and can be atypical. The severity of clinical symptoms is significantly associated with the size of hemorrhage, degree of hematoma, and length of onset time. It is most commonly encountered in the lower cervical and upper thoracic spinal levels, although it may occur across the whole spine. Favorable neurological and functional outcomes can be achieved with early, accurate diagnosis, and emergent surgical treatment. In addition, early and individualized rehabilitation program can prevent long-term disability in SSEH patients.

In this report, we present a male case who developed quadriaparesis during acute coronary syndrome (ACS) treatment and underwent emergency surgery and received early rehabilitation to prevent neurological sequelae.

CASE REPORT

A 65-year-old male patient was admitted to our hospital with chest pain in the coronary intensive care unit. Upon the detection of instable angina pectoris,
60 mg low-molecular-weight heparin (LMWH) and 300 mg clopidogrel were initiated. The patient was on antihypertensive treatment for 15 years due to essential hypertension and he also used acetylsalicylic acid 100 mg/day. The patient did not have any a recent head or spinal trauma and surgical treatment, and he did not smoke.

He experienced sudden-onset severe neck pain and occipital headache on the third day of ACS treatment and felt muscle weakness and numbness in bilateral upper and lower extremities about 24 h after the onset of pain. On his physical examination, the patient was alert, orientated, and afebrile with a blood pressure of 140/90 mmHg. The muscle strength of upper extremities was 2/5 proximally and 1/5 distally, while that of the lower extremities was 0/5 proximally and distally. There was anesthesia below the C4 dermatome. Also, no deep tendon reflex could be elicited. The rectal examination revealed reduced anal sphincter tone and anesthesia in the perineal region. Routine laboratory and coagulation parameters (prothrombin time and activated partial thromboplastin time, and the international normalized ratio) were within normal limits. Neurology and neurosurgery consultations were requested, and urgent spinal magnetic resonance imaging (MRI) was performed. The MRI showed a space occupying formation extending from the level of C4 to T1 in the cervical spine which did not cause abnormal signal changes in the spinal cord and which was isointense in the T1-weighted images and heterogeneous signal intensity in the T2-weighted images. Spinal cord injury was assessed as Grade A complete injury according to the American Spinal Injury Association (ASIA) Impairment Scale. The patient underwent emergency decompressive laminectomy and hematoma evacuation with the diagnosis of SSEH approximately 40 h after the onset of symptoms. On the postoperative Day 1, no neurological recovery was noted, while the muscle strength of the lower extremities was 1/5 after two weeks. The range of motion exercises for upper and lower extremities, muscle strength enhance training, seat balancing training, transfer training (from the bed to a wheelchair), and respiratory function training were given to the patient as the early rehabilitation program. Also, clean intermittent self-catheterization was initiated for him in whom urinary retention persisted. After 60 sessions of rehabilitation program, neurological recovery was achieved in the upper (proximal 3/5, distal 1/5) and lower (proximal 2/5, distal 1/5) extremities. A written informed consent was obtained from the patient.

**DISCUSSION**

The sole use or various combinations of anticoagulant and antiplatelet drugs used for the treatment of cardiovascular and cerebrovascular diseases are one of the most common factors regarding the development of SSEH. Antiplatelet agents associated SSEH is quite rare, whereas
25 to 70% of SSEH patients were reported to have the history of anticoagulation use. Unlike anticoagulants, the risk of major hemorrhage is also lower for antiplatelet agents. However, Harker et al. reported that the incidence of hemorrhagic events did not differ significantly among the groups receiving clopidogrel and acetylsalicylic acid treatments. The patients receiving a combination of acetylsalicylic acid and clopidogrel were also demonstrated to have a higher prevalence of major hemorrhage than those treated with only clopidogrel or acetylsalicylic acid. In addition, the risk of major hemorrhagic complications increases, when concurrent antiplatelet agents are used in combination with LMWH. Our case used acetylsalicylic acid 100 mg/day and was on antihypertensive treatment for 15 years due to essential hypertension treatment. It can be speculated that adding LMWH and clopidogrel to the treatment due to ACS may have had a direct impact on the development of SSEH.

The pathophysiology of SSEH has not been completely understood yet, and it is still controversial whether the origin of epidural hematomas is arterial or venous. Since spinal epidural venous plexuses which do not contain any valves drain to abdominal and thoracic venous systems, vigorous physical activity enough to cause changes in the abdominal or thoracic pressure may lead to rupture by causing a sudden increase in the spinal venous pressure. However, since cervical epidural venous pressure is lower than the intrathecal pressure, venous hemorrhage cannot cause compression in the dural sac. Therefore, particularly cervical SSEHs may be considered to originate from epidural arteries. The fact that our patient had a hematoma in the cervical region, that he had a history of hypertension, and that there was no severe physical strain or trauma prior to his symptoms suggests that the hematoma may be of arterial origin.

Although SSEH has a characteristic clinical presentation, its early and accurate diagnosis still remains a challenge due to its rare prevalence and usually varying and atypical initial symptoms. In case of sudden spinal pain accompanied by a neurological deficit, a potential hemorrhagic disease of the spinal cord should be considered. The differential diagnosis highly depends on a pertinent clinical and radiological correlation. A heterogeneous signal intensity or hyperintensity appearance on T2-weighted MRIs and homogeneous hypointensity or isointense appearance on T1-weighted images are typical for SSEH. Early diagnosis plays an important role for the prognosis of SSEH and probability of initiating surgical treatment.

The prognosis of SSEH can be associated with the size of hematoma, severity of preoperative paresis, and time interval between the onset of symptom and surgical treatment. Groen reported that the size of hematoma was larger in patients with conservative follow-up treatment, compared to patients treated surgically, suggesting that hematoma size does not affect prognosis and should not be used in treatment selection. On the contrary, examining SSEH patients treated surgically, Liao et al. found that the large size of hematoma and a hematoma extending over two or more segments caused an increase in postoperative relapse rate and resulted in worse functional recovery. The early surgical treatment of SSEHs, particularly in the cervical and thoracic region, yielded more favorable neurological and functional outcomes in patients with incomplete spinal cord injury than those with complete injury. Previous studies indicated that the surgical treatment outcomes of SSEH were inversely associated with the time interval between the onset of symptoms and surgical treatment. Shin et al. suggested that more favorable neurological outcomes were observed in patients operated within 12 h. In the study by Groen and van Alphen, evaluating 330 patients with SSEH, surgical treatment produced more favorable outcomes, when performed before 36 h in patients with complete injury and before 48 h in patients with incomplete injury. Although the clinical presentation of our case was typical, administering surgical intervention after the emergence of neurological deficits that may affect long-term outcomes (>36 h), the presence of severe paresis (ASIA A) and a hematoma extending over two or more segments (four segments) might have affected prognosis adversely and caused a limited postoperative neurological and functional recovery in our case.

The gold standard treatment of SSEH is urgent decompressive laminectomy and hematoma evacuation. Early decompressive surgical treatment was shown to affect neurological recovery and functional outcomes positively. According to Liao et al., complete recovery in the postoperative first year was 88.9% in patients with incomplete injury, while it was only 37.5% in those with complete injury. However, conservative treatment may be considered for patients with rapid spontaneous recovery of neurological deficits in the early period or patients with mild neurological deficits (ASIA D or E). It can also be used in case of advanced cardiovascular
disease, progressive and irreversible spinal cord injury, or for high-risk patients for hemorrhagic tendency.

In conclusion, non-traumatic hematomas of the spinal column are rare and severe, and they can cause severe neurological sequelae. Spontaneous spinal epidural hematoma should be suspected in case of spinal pain accompanied by motor or sensorial deficit in any patient receiving anticoagulation and/or antiplatelet therapy. Early diagnosis and urgent surgical decompression are warranted for reducing neurological sequelae.

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