

# Intracranial Vasospasm without Intracranial Hemorrhage due to Acute Spontaneous Spinal Subdural Hematoma

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Spontaneous spinal subdural hematoma (SDH) is very rare. Furthermore, intracranial vasospasm (ICVS) associated with spinal hemorrhage has been very rarely reported. We present an ICVS case without intracranial hemorrhage following SDH. A 41-year-old woman was admitted to our hospital with a complaint of severe headache. Multiple intracranial vasospasms were noted on a brain CT angiogram and transfemoral cerebral angiography. However, intracranial hemorrhage was not revealed by brain MRI or CT. On day 3 after admission, weakness of both legs and urinary incontinence developed. Spine MRI showed C7~T6 spinal cord compression due to hyperacute stage of SDH. After hematoma evacuation, her symptoms gradually improved. We suggest that spinal cord evaluation should be considered in patients with headache who have ICVS, although intracranial hemorrhage would not be visible in brain images.

**Key words:** Spinal subdural hematoma, Subarachnoid hemorrhage, Intracranial vasospasm, Headache

## INTRODUCTION

Intracranial vasospasm (ICVS) can be caused by any situation that brings about bleeding into the cerebral subarachnoid space, such as traumatic subarachnoid hemorrhage (SAH), cerebral tumoral bleeding, or rupture of vascular malformations, but the rupture of cerebral aneurysms within the basal cistern is most commonly associated with ICVS [1]. Moreover, migraine, taking vasoactive drugs (cocaine, pseudoephedrine, immunosuppressants, or

selective serotonin reuptake inhibitors), or the postpartum period can also cause ICVS and tend to induce vasospasm most often without intracranial bleeding [2]. Because ICVS leads to cerebral ischemia that can cause devastating neurological deterioration in some cases, it is important that ICVS be detected and its cause be identified as early as possible [1]. Acute spontaneous spinal subdural hematoma (SDH) is very rare. However, it often results in serious complications, hence appropriate therapeutic approaches and rapid diagnosis are needed [3, 4]. ICVS associated with spontaneous spinal SDH has also been reported very rarely [4]. Here, we firstly report a case of ICVS without intracranial hemorrhage that was caused by acute spontaneous spinal SDH.

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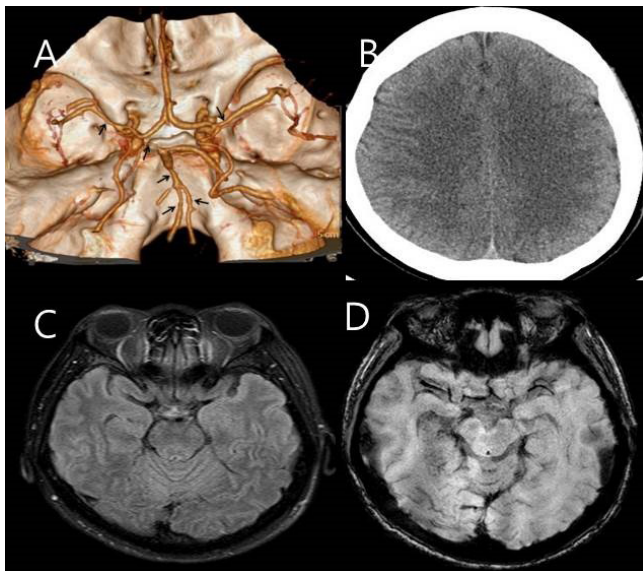
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## CASE

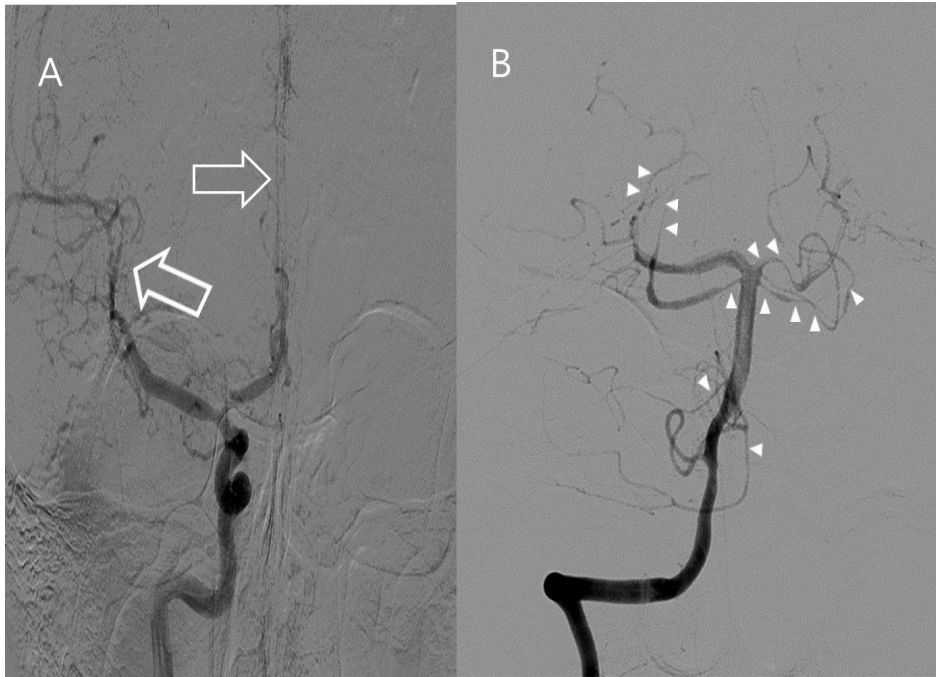
A 41-year-old woman was admitted to our hospital with a complaint of severe headache. She had headache for 3 years and expressed it as throbbing pain in both temporal regions accompanied by nausea. This pain was exacerbated by physical activity and under sunlight. Symptoms tended to occur once in 1~2 months, lasted for 1~3 days and then disappeared. A week before admission to our hospital, her headache symptoms were different from those in the past. She felt a twinge at the right posterior neck for the first time, and subsequently experienced severe pain in the entire head, but no nausea, vomiting, or fever. She was admitted to the neurology department of an outside hospital and underwent computerized tomography (CT) angiography 2 days after the headache developed; no bleeding was noticed yet multifocal vasospasms of intracranial arteries were revealed (Fig. 1A, B). She was diagnosed with status migrainosus; hence, steroid pulse therapy with oral beta-blockers and non-steroidal anti-inflammatory drugs were given to relieve headache. However, her symptoms did not improve but rather worsened. Several hours before admission to our hospital, she started complaining of nausea and vomiting as well as very sharp tearing pain in the neck and back. According to the analysis of a cerebrospinal fluid (CSF) specimen that was obtained via lumbar puncture in an outside hospital 3 days after symptom onset, the levels of white blood

cells (WBC), protein, and glucose were 4/ $\mu$ l, 62 mg/dl, and 46 mg/dl, respectively. Other CSF profiles were not provided. She had a history of hypertension and her blood pressure was well controlled with regular antihypertensive medication. She denied any history of auto-immune or cerebrovascular diseases. There was also no special family history of migraine or autoimmune disease. On admission to our hospital, her blood pressure was 195/114 mmHg; pulse, 60 beats/min; respiration, 20 breaths /min; and body temperature, 36.6°C. Mental status was alert and oriented. Cranial nerve examination and motor and sensory functions were normal. No pathologic reflexes were found, yet neck stiffness was suspected. According to the routine blood tests, the only abnormality was that the WBC count was increased to 18000/ $\mu$ l. In the blood coagulation test, prothrombin time–international normalized ratio and activated partial thromboplastin time were 0.99 s (normal range, 0.00~1.20 s) and 28 s (normal range, 20.0~36.0 s), respectively; both values were within the normal range. Aortic CT angiography revealed no evidence of aortic dissection. We started steroid pulse and mannitolization therapy. On day 2 after admission, her headache was alleviated, yet pain in the neck and back rapidly worsened and subsequently spread to the whole spine. Additionally, marked neck stiffness was observed. On day 3 after admission, brain magnetic resonance imaging (MRI) showed no intracranial hemorrhage, such as SAH (Fig. 1C, D). On the same day, transfemoral cerebral angiography (TFCA) revealed multiple ICVSs (Fig. 2A, B). Two hours after TFCA, weakness of both legs [Medical Research Council (MRC) grade 0 or I], bladder distension, and sensory deficit below both nipples developed. On spinal cord MRI, we found spinal cord compression by SDH, which was located from the ventral lower cervical to mid-thoracic level (C7 to T6) (Fig. 3). In subsequent CSF examinations, the red blood cell count was 340000/ $\mu$ l and WBC count was 30/ $\mu$ l (polymorphonuclear leukocytes, 59%; lymphocytes, 29%; and mononuclear cells, 12%), CSF was red, and no color change was observed in the three-tube test. Emergent total laminectomy was performed through the posterior approach, which was started 7 h after paraplegia. While incising the dura, we found that the spinal cord was swollen and large hematoma was present in the subdural and subarachnoid spaces. The hematoma was evacuated and no vascular malformation was found. After surgery, pain was relieved. On postoperative day 5, sensory symptoms and muscle power of the left leg (MRC grade II) partially improved.

Upon the request by the patient's guardian, the patient was transferred to another hospital on postoperative day 6. Three days after the transfer, the patient had weakness and alien hand syndrome of the left arm. Brain MRI showed bilateral multifocal acute infarction of the corpus callosum; the right side of the lesion



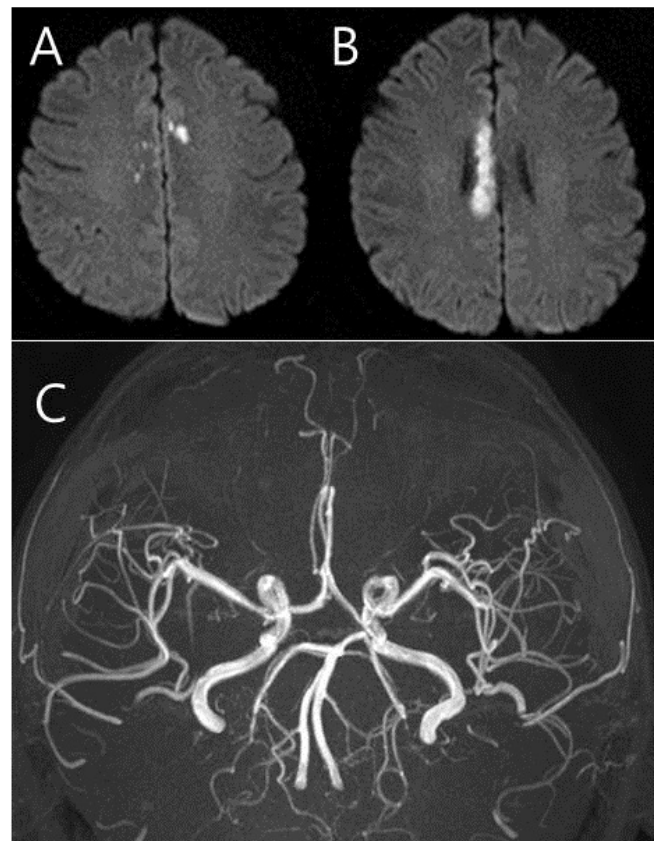
**Fig. 1.** Brain CT angiography, CT and MRI. (A, B) Brain CT angiography 2 days after symptom onset shows multifocal intracranial vasospasms (arrows) without intracranial hemorrhage. (C, D) Brain fluid-attenuated inversion recovery (C) and susceptibility-weighted (D) imaging 9 days after symptom onset also do not show intracranial hemorrhage.



**Fig. 2.** Transfemoral cerebral angiography 9 days after symptom onset. Multifocal vasospasms are shown in the left carotid (A, arrows) and left vertebral (B, arrowheads) angiograms.



**Fig. 3.** T2-weighted sagittal MRI of spinal cord shows an acute hematoma (black arrows) at the level of C7~T6 segments with cord compression. There is also mildly increased T2 signal intensity within T1 and T2 segments. T2 axial MRI at the T2 spine level shows a hematoma (white arrows) within the extramedullary and intradural space, which means that the lesion is of the subdural origin (left lower corner box).



**Fig. 4.** (A, B) Brain diffusion MRIs 17 days after symptom onset. Acute bilateral multifocal infarctions are shown in the corpus callosum area. (C) MR angiography reveals no vasospasm in any of the intracranial arteries about 1 year after symptom onset.



was larger than the left side (Fig. 4A, B). On CT angiography performed at the other hospital at 8 days after the transfer from our hospital, more exacerbated vasospasms were found in the anterior, posterior, and middle cerebral arteries than initial CT angiography after the headache onset. Upon medical treatment, symptoms in the left arm gradually improved. Four months after discharge from our hospital, she visited our out-patient clinic and no longer had any pain including headache. She did not complain of urinary symptoms yet complained of a tingling feeling in both legs. The muscle power of both legs and the left arm was approximately MRC grade IV, while the power of the right arm was MRC grade V, which represented considerable improvements in comparison with those parameters at the time of discharge from our hospital. Alien hand syndrome was also no longer observed.

About 1 year after symptom onset, muscle power recovered to an almost normal level in both legs and arms. Follow-up magnetic resonance angiography revealed no vasospasms in any of the intracranial arteries (Fig. 4C).

## DISCUSSION

As opposed to the brain, the subdural cavity of the spinal cord lacks a bridging vein; thus, the mechanisms of spinal SDH development are still unclear. Although subdural reticular anastomosis can be damaged during lumbar puncture, it is very unlikely to explain SDH, given the size of the vessels [5]. Rader [6] suggested that SDH might be caused by indirect force delivered to the vessels in the spinal cord. A sudden rise in intra-abdominal or intra-thoracic pressure is known to increase the inner pressure in subdural and subarachnoid vessels, thereby subsequently resulting in their rupture.

Calhoun and Boop [7] suggested that bleeding in the subarachnoid cavity results in SDH. The radiculomedullary arteries and veins in the subarachnoid cavity are large enough to cause bleeding. In the early stage of bleeding, a blood clot may not form because of the redistribution and dilution of the blood by the CSF flow. As bleeding progresses, clots of considerable size can form, which may be sufficient to block the CSF flow; CSF flow stagnation can increase the buildup of clots in the subarachnoid space, which eventually leads to a rupture of the arachnoid membrane, thereby causing SDH. In our patient, pain started initially in the posterior neck, then spread to the entire head, and then to the whole spine. After the development of these symptoms, transverse myelopathy symptoms (paraplegia, bladder distension, and sensory deficit below both nipples) developed. Furthermore, the fact that bleeding was found in the subarachnoid space during surgery indicates that SAH at the upper thoracic level had spread

out to the intracranial cavity and caused ICVSs, which presumably led to headache; with further progression of the subarachnoid hemorrhage at the thoracic level, the CSF flow may have been hindered, in turn resulting in clot enlargement. Consequently, these clots may have led to SDH development and subsequently induced spinal cord compression. Taking into account these speculations, the hypothesis of Calhoun and Boop appears to more reasonably explain the mechanisms of spinal SDH than hypotheses of the others in this case.

Intracranial vasospasm can develop 3~14 days after aneurysmal SAH [8]. The amount of bleeding in the cistern would be closely related to vasospasm symptoms [9, 10]. Interestingly, our patient exhibited not only intracranial vasospasm but also cerebral infarction. However, SAH was not observed on brain MRI or CT. In a previous report [11], the rate of SAH diagnosis varied depending on the time from symptom onset to CT imaging. The delay was associated with the lower sensitivity of SAH diagnosis (98~100% within the first 12 h, 93% within 24 h, 57~85% after 6 days). On spinal cord MRI of our patient, the hematoma was located longitudinally. The lesion above the T2 spine segment was thinner and showed lower signal intensity. Additionally, radiologic modalities such as MRI or CT are known not to be able to differentiate between subarachnoid and subdural hemorrhage [8]. Therefore, we may have not detected SAH because 1) the patient underwent brain imaging too late (initial brain imaging was performed 2 days after symptom onset) or 2) the spread of the hemorrhage into the cranial cavity was limited.

In a recent report, Shakur and Farhat [8] presented a case similar to ours. However, unlike in our case, they confirmed cortical SAH and ventricular hemorrhage by CT. Major symptoms of acute spinal SDH include sudden-onset pain in the back and neck, radicular pain, as well as myelopathy symptoms (e.g., paraplegia, paresthesia, and dysuresia) [10, 12]. The risk factors for spinal SDH include the use of anticoagulants, coagulopathy, vascular malformations, spinal infection, and tumors [13]. Lumbar puncture is also associated with spinal SDH [14]. The characteristics of previous headache that our patient had for 3 years seemed to indicate that she suffered from migraine. As reported previously, ICVS can be seen in patients with migraine [2]. Our patient initially presented with headache, whereas myelopathy symptoms developed later, after the first lumbar puncture. However, the characteristics of her headache at admission to the outside hospital were different from previous symptom of headache, and spinal SDH was located from the lower cervical to mid-thoracic segment. If spinal SDH had been caused by the lumbar puncture, spinal SDH would have been found around the lumbar area. Thus, we do not believe that the lumbar puncture caused spinal SDH.

According to Domenicucci et al. [3], in 15% out of 106 cases, the causes of non-traumatic subdural spinal hematomas were unknown. Our patient did not have any risk factors for hemorrhage and complained of severe headache in the early stage; imaging revealed no hemorrhage (e.g., SAH), while vasospasms were clearly established. It has been suggested that emergency surgical decompression is critical to determine prognosis in patients with acute spinal SDH [3]. Therefore, it is important to note that there is a possibility of acute spontaneous spinal SDH in patients with headache who have ICVS despite no intracranial hemorrhage detected by brain imaging.

#### ACKNOWLEDGEMENTS

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