Accidental subarachnoid hemorrhage due to undiscovered cerebral venous sinus thrombosis during cesarean section

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INTRODUCTION

Cerebral venous sinus thrombosis (CVST), a rare cerebrovascular event, occurs in approximately 5 out of 1,000,000 people [1]. It is frequently associated with pregnancy and puerperium, as well as with oral contraceptive use, coagulopathies, and infection [2]. Although pregnancy-associated CVST is not common, the risk is greatly increased during the last trimester of pregnancy and puerperium [3]. Typical clinical manifestations of CVST include acute headache, blurry vision, focal neurologic deficits, altered level of consciousness, and seizures [4]. This report describes a woman who experienced a severe intraoperative headache due to a subarachnoid hemorrhage (SAH) as the first intraoperative symptom of CVST while undergoing a cesarean section.

CASE REPORT

A primiparous pregnant woman, aged 36 years, presented to the emergency room at 37 weeks and 5 days of gestation with persistent labor pains and vaginal bleeding that had started on the day of presentation. The patient had previously been diagnosed with gestational diabetes and indeterminate arrhythmia, but the results of preoperative laborato-
ry tests, chest radiography, and electrocardiography (EKG) were normal. An emergency cesarean section was deemed necessary. Initially, her vital signs in the operating room were stable, and she was administered spinal anesthesia at the lumbar 4-5 level with a 25-gauge Quincke spinal needle. Subsequently, her systolic blood pressure dropped to 70 mmHg and her pulse rate to 56 beats/min, and she began to experience nausea. She was injected sequentially with 5 mg of ephedrine and 100 μg of phenylephrine, but she experienced an abrupt and severe right-sided frontal headache, accompanied by right eye pain. Her blood pressure suddenly rose to 184/115 mmHg and her pulse rate to 138 beats/min, with EKG at the time of birth showing about 4.0 mm of ST depression. She was injected with 5 mg esmolol to manage her tachycardia, with her blood pressure being 174/76 mmHg after 5 min. Subsequently, her blood pressure decreased to 132/89 mmHg, but her heart rate remained fast at 137 beats/min.

The patient remained alert with no neurologic deficits. Arterial blood gas analysis and cardiac marker tests were performed for differential diagnosis. General anesthesia was induced, because the patient showed persistent and severe body movements, making the operation much more difficult to perform. After maintaining appropriate general anesthetic depth, her vital signs and ST changes on the EKG gradually returned to normal, and the arterial blood gas and laboratory tests showed no abnormalities. The surgery was completed without other complications.

After emergence from anesthesia, the patient showed muscle strength of grade 2-3 of 5 and limited movement of her left arm and leg. Computed tomography (CT) of the brain showed an SAH in the right frontal sulci with surrounding vasogenic edema in the right frontal lobe and subdural hemorrhage in the right lateral convexity, along with slight midline shifting to the left (Fig. 1). Based on the patient’s age and medical history, a transfemoral cerebral an-
giography (TFCA) was performed to rule out arteriovenous malformation. The TFCA showed no significant abnormality around the hemorrhagic lesion. Over time, however, the patient’s level of consciousness decreased to a drowsy state and her left side weakness worsened to grade 0/5 with mild left facial palsy. To determine the cause of the hemorrhage, brain magnetic resonance imaging and magnetic resonance venography were performed. These imaging modalities showed stenosis in the anterior superior sagittal sinus (Fig. 2). Because of the risk of postpartum hemorrhage, anticoagulation treatment was not started immediately. Rather, she was followed-up by brain CT to determine if the bleeding worsened.

Despite efforts to control intracranial pressure, her level of consciousness decreased to stupor and brain CT on postoperative day (POD) 5 showed progressive cerebral edema. Following decompressive craniectomy, the patient became alert and stable, and she was started on anticoagulation therapy with Fraxiparine for 10 days. The patient has been undergoing global rehabilitation for left hemiplegia (G1/G1), left facial palsy, and mild dysarthria, and she continues to receive anticoagulation therapy with warfarin. Although her serum concentration of antithrombin III was not determined preoperatively, postoperative examination on POD 3 showed that her antithrombin III level was 67% of normal.

DISCUSSION

Early diagnosis of CVST in this patient was very difficult because of the atypical characteristics of the headache that occurred during the cesarean section. At first, we suspected preeclampsia or postdural puncture headache rather than intracranial hemorrhage, because the former are more frequent causes of intraoperative headache in pregnant women under spinal anesthesia. Headache appears in almost 90% of patients with CVST, whereas severe and sudden headache is typical in patients with SAH [5]. In addition, SAH secondary to CVST is extremely rare [6], delaying the diagnosis of SAH due to CVST until the patient showed reduced motor function after emergence from anesthesia, indicating the necessity of neurologic evaluation and suggesting a diagnosis of intracerebral hemorrhage.

The onset and origin of CVST in this patient remain unknown. Although CVST in a hypercoagulable state was thought to be caused by hyperhomocysteinemia [7] or antithrombin III deficiency [8], no case reports to date have described CVST in a young pregnant woman with no significant medical conditions. Her postoperative antithrombin III level was 67%, lower than normal. Antithrombin III concentrations normally decrease during pregnancy, being 30% of baseline during the postpartum period [9]. Pregnancy has been found to induce a hypercoagulable state [10], which persists for several weeks during the postpartum period [11]. Pregnancy-related cerebral venous thrombosis (CVT) has also been reported, with the risk of peripartum CVT increasing with maternal age and cesarean delivery, as well as in women with hypertension, infections, and excessive vomiting during pregnancy [12].

Several mechanisms may explain the condition experienced by this patient. One possible mechanism is the rupture of venous parenchymal hemorrhagic infarcts into the subarachnoid space [13]. The high blood pressure experienced by this patient immediately after induction of spinal anesthesia may represent an effort to counteract the increased intracranial hypertension caused by venous thrombosis. Another possible mechanism involves the rupture of dilated, valveless, thin-walled, bridging subarachnoid cortical veins lacking smooth muscle fibers, occurring subsequent to venous hypertension [14]. Moreover, the pattern of distribution of SAH associated with CVST differs from that of SAH of arterial origin. For example, SAH located at the cerebral convexity and sparing the basal cisterns and skull base may be indicative of SAH due to CVST [15]. Under these conditions, a subdural hematoma (SDH) in patients with CVST would be caused by rupture of bridging veins that connect superficial cerebral veins and the dural venous sinuses by back pressure changes in the venous channels proximal to the thrombosed cerebral venous sinuses. Alterations in consciousness level and motor function would therefore be due to increased cerebral pressure caused by simultaneous SDH and SAH.

Spinal anesthesia and the delivery process during cesarean section are accompanied by major changes in hemodynamics and surgical stress. Brain hemorrhage in this patient with CVST during pregnancy likely resulted from sudden changes in hemodynamics and surgical stress during the course of anesthesia induction and surgery. High blood pressure after delivery may be regarded as a compensatory mechanism
due to an increase in brain pressure, severe headache, and/or discomfort caused by pressing on the upper abdomen during delivery. The Cushing phenomenon, a mechanism that compensates for increased brain pressure, was less likely, as it is accompanied by bradycardia.

The management of CVST usually starts with the administration of systemic anticoagulants such as heparin or low molecular weight heparin because of their efficacy, safety, and feasibility. Some patients with SAH have also been safely treated with anticoagulation therapy [16]. However, major bleeding, the most severe potential complication of any form of heparin therapy, may occur. Anticoagulation therapy was initiated very carefully in this patient, especially as she had just undergone major surgery. Moreover, both surgical wound bleeding and intracranial bleeding owing to the rupture were persistent, making tight blood pressure control and close neurological monitoring the best option for this patient. Anticoagulation therapy may not be optimal for all patients, indicating a need for individualized management. Neurosurgical intervention is dependent on the amount of hemorrhage, the degree of midline shift, and the presence of symptoms [17]. However, consultation with a neurosurgeon revealed that the risk of surgical intervention was greater than the benefit.

In conclusion, anesthesiologists should be aware of the possibility of SAH due to CVST in patients who complain of nonspecific headache during pregnancy or delivery. Early diagnosis and appropriate management are essential for favorable outcomes.

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CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Ferro JM, Aguiar de Sousa D. Cerebral venous thrombosis: an update. Curr Neurol Neurosci Rep 2019:19:74.

2. Meng R, Ji X, Wang X, Ding Y. The etiologies of new cases of cerebral venous sinus thrombosis reported in the past year. Intractable Rare Dis Res 2012;1:23-6.

3. Ferro JM, Canhão P. Cerebral venous sinus thrombosis: update on diagnosis and management. Curr Cardiol Rep 2014;16:523.

4. Liang Zw, Gao WL, Feng LM. Clinical characteristics and prognosis of cerebral venous thrombosis in Chinese women during pregnancy and puerperium. Sci Rep 2017;7:43866.

5. Einhäupl K, Stam J, Bousser MG, De Bruijn SF, Ferro JM, Martinelli I, et al. EFNS guideline on the treatment of cerebral venous and sinus thrombosis in adult patients. Eur J Neurol 2010;17:1229-35.

6. Sahin N, Solak A, Gene B, Bilgie N. Cerebral venous thrombosis as a rare cause of subarachnoid hemorrhage: case report and literature review. Clin Imaging 2014;38:373-9.

7. Arévalo-Lorido JC, Carretero-Gómez J. Cerebral venous thrombosis with subarachnoid hemorrhage: a case report. Clin Med Res 2015;13:40-3.

8. Elias N, MuraiRajamany K, Adenan NAM. A case report of fatal outcome of cerebral venous sinus thrombosis with cerebral hemorrhage during early pregnancy secondary to antithrombin III deficiency. Int J Womens Health Reprod Sci 2018;6:220-2.

9. James AH, Rhee E, Thames B, Philipp CS. Characterization of antithrombin levels in pregnancy. Thromb Res 2014;134:648-51.

10. Yamamoto J, Kakeda S, Takahashi M, Idei M, Nakano Y, Soejima Y, et al. Severe subarachnoid hemorrhage associated with cerebral venous thrombosis in early pregnancy: a case report. J Emerg Med 2013;45:849-55.

11. Cole B, Criddle LM. A case of postpartum cerebral venous thrombosis. J Neurosci Nurs 2006;38:350-3.

12. Saposnik G, Barinagarrementeria F, Brown RD Jr, Bushnell CD, Cucchiara B, Cushman M, et al. Diagnosis and management of cerebral venous thrombosis: a statement for healthcare professionals from the American Heart Association/American Stroke Association. Stroke 2011;42:1158-92.

13. Kato Y, Takeda H, Furuya D, Nagoya H, Deguchi I, Fukuoka T, et al. Subarachnoid hemorrhage as the initial presentation of cerebral venous thrombosis. Intern Med 2010;49:67-70.

14. Cuvinciuc V, Viguier A, Calviere L, Raposo N, Larrue V, Cognard C, et al. Isolated acute nontraumatic cortical subarachnoid hemorrhage. AJNR Am J Neuroradiol 2010;31:355-62.

15. Akins PT, Axelrod YK, Ji C, Ciporen JN, Arshad ST, Hawk MW, et al. Cerebral venous sinus thrombosis complicated by subdural hematomas: Case series and literature review. Surg Neurol Int 2013;4:85.

16. Rodallec MH, Krainik A, Feydy A, Hélias A, Colombani JM,
Jullés MC, et al. Cerebral venous thrombosis and multidetector CT angiography: tips and tricks. Radiographics 2006;26 Suppl 1:S5-18; discussion S42-3.

17. Liu Y, Li K, Huang Y, Sun J, Gao X. Treatment of the superior sagittal sinus and transverse sinus thrombosis associated with intracranial hemorrhage with the mechanical thrombectomy and thrombolytics: case report. Medicine (Baltimore) 2017;96:e9038.