

**Case
Report**

Acute Subdural Hematoma Following Thoracoabdominal Aortic Repair in a Patient with Marfan Syndrome: A Rare Complication of Cerebrospinal Fluid Drainage

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Cerebrospinal fluid (CSF) drainage is a routinely used adjunct in operation of thoracoabdominal aortic aneurysm (TAAA), which may reduce the incidence of perioperative paraplegia by improving spinal cord perfusion. Neurological complications of CSF drainage have been reported, possibly due to excessive CSF drainage, and acute subdural hematoma (SDH) in particular may lead to catastrophic complications. We present a rare case of acute SDH due to CSF drainage that was not excessive, after TAAA repair in a patient with Marfan syndrome, who recovered without invasive treatment.

Keywords: CSF drainage, acute subdural hematoma, thoracoabdominal aortic aneurysm

Introduction

Cerebrospinal fluid (CSF) drainage is hypothesized to decrease the intrathecal pressure, thus increasing medullary perfusion and minimizing potential spinal cord ischemia.^{1–3)} Although CSF drainage has become a standard method to reduce the risk of paraplegia in thoracoabdominal aortic aneurysm (TAAA) repair, acute subdural hematoma (SDH) associated with the placement of lumbar CSF drainage catheters has been reported.^{4–6)} In particular, it has been reported that a larger volume of CSF drainage is a significant risk factor for SDH.⁷⁾ Herein, we present a rare case of acute SDH in a patient who did not undergo excessive CSF drainage and recovered without invasive treatment.

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Case Report

A 29-year-old woman with Marfan syndrome was hospitalized for elective repair of thoracoabdominal aortic dissection. Nine months before her admission, she had undergone replacement of an aortic root and ascending aorta for Stanford type A acute aortic dissection. Preoperative laboratory parameters including those of coagulation studies were within normal ranges. There were no findings of intracranial abnormalities on preoperative brain computed tomography (CT). One day before the operation, a spinal fluid drainage catheter (Silascon™, Kaneka Corp., Tokyo, Japan) was inserted into the L3-L4 vertebral interspace with the patient in the right lateral decubitus position, and the catheter was advanced 10 cm after it had entered the dura. Blood was not aspirated from the needle during drain insertion. The CSF drainage catheter was connected to a closed collection system which was stopped until surgery was begun. The elapsed time between lumbar CSF catheter insertion and systemic heparinization was approximately 24 hours. Continuous, unlimited CSF drainage was performed during the operation and 24 hours, postoperatively. During that time, the CSF pressure remained constant at approximately 10 mm Hg. Spinal fluid pressure (SFP) was measured during and

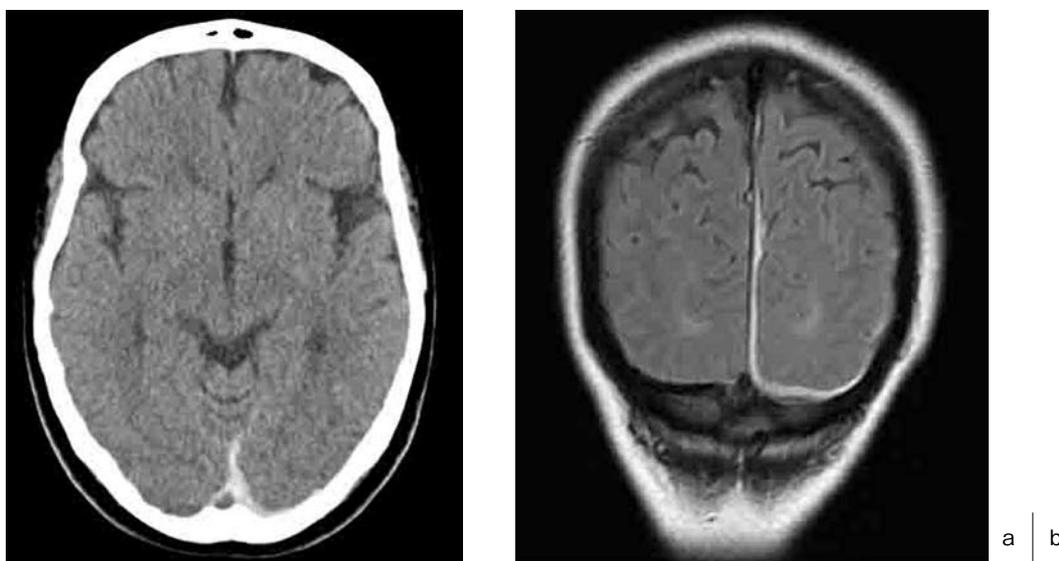


Fig. 1 a: Brain computed tomography of the brain on postoperative day 2 reveals a small subdural hematoma at the superior sagittal sinus region.
 b: Brain magnetic resonance imaging of the brain on the postoperative day 2 reveals a small subdural hematoma at the superior sagittal sinus region.

after the operation, until the patient was awake and able to perform a normal leg lift. The aortic aneurysm was replaced with the use of a 24 mm vascular graft (Coselli Thoracoabdominal Graft™, Termo Corp., Tokyo, Japan), which extended from 2 cm distal to the origin of the left subclavian artery to the terminal abdominal aorta. The intercostals (TH8-12), celiac, superior mesenteric, renal and inferior mesenteric arteries were reimplemented. The operation technique used standard methods previously detailed by many authors,^{1,2,8)} including permissive mild hypothermia (32°C–34°C, nasopharyngeal), warm blood perfusion of celiac and superior mesenteric arteries, perfusion of renal arteries with 4°C crystalloid solution, aggressive reattachment of segmental arteries, especially between Th8 and Th12, normal heparinization (3 mg/kg) and a distal aortic perfusion technique.²⁾ Distal aortic perfusion was used to maintain arterial pressure at about 60 mm Hg from the distal site of the cross clamp by femoro-femoral bypass. The procedure was uneventful, and the operating time was 425 min. The total amount of CSF drainage during the operation was 85 mL. The patient was awake a few hours after the operation in the ICU and extubated the next morning. Drainage of CSF was stopped 24 hours after the operation, and the total amount of CSF leakage was 125 mL. On postoperative day 2, the patient suffered a severe headache; therefore, brain computed tomography (CT) and brain magnetic resonance imaging (MRI) were performed (**Fig. 1a** and **1b**). A high

density shadow was found at the superior sagittal sinus region on the brain CT, and a high intensity area was found in the subdural space of the left lateral lobe and posterior lobe by MRI (FLAIR). After consultations between the neurologist and radiologist, the patient was diagnosed with acute subdural hematoma (**Fig. 1a** and **1b**).

There were no neurological deficits accompanying the headache. Treatment of the patient was conservative without craniectomy or ventricular drainage. One week after the operation, the headache was gone, and the patient was discharged 1 month after the operation. The follow-up brain CT showed an improvement in the lesion (**Fig. 2**).

Discussion

In a prospective randomized clinical trial, Coselli et al. showed that perioperative CSF drainage up to 10 mmHg reduced the rate of paraplegia after repair of Crawford extent I and II TAAAs.¹⁾ In contrast, several other authors have reported that CSF drainage can be associated with significant complications.^{4–6)} Weaver et al. reported that the incidence of SDH was 3.2% after CSF catheter drainage in thoracic aortic aneurysm repairs⁴⁾ and Dardik et al. reported that a larger volume of CSF (mean 178 mL) drainage in the perioperative period significantly increased risk of SDH.^{5,7)} However, SDH occurred in our case despite a relatively low total drainage volume of only



Fig. 2 Follow up brain computed tomography showed an improvement in the lesion (two months after the operation).

125 mL. The mechanism by which SDH coincides with CSF drainage is not completely understood. Previous studies have shown that subdural hematoma may result from stretching and tearing of the bridging vein between the dura and cerebral hemispheres in cases of excessive CSF drainage or shrinkage of the brain due to dehydration.^{9,10} In our case, we suspected that sudden drops in CSF pressure at the insertion of CSF drainage, and not excessive CSF drainage, might be the cause of intracranial hypotension and rupture of the cortical veins, however, the pressure of CSF was continued 10 mmHg (13.5 cm H₂O) during and after the operation. Although the bleeding may have occurred just after the insertion, the hemorrhagic space was only limited at very narrow space of sagittal sinus region because CSF drainage tube that was closed just after insertion before 24 hours of systemic heparinization and the pressure of CSF was continuously kept at 10 mmHg (13.5 cm H₂O) during the operation.

However, it is unclear whether the subarachnoid system in patients with Marfan syndrome is especially vulnerable to aspiration pressure changes.

Many cases of SDH are life threatening, with almost 60% mortality reported due to massive bleeding in the brain.¹⁰ Usually CSF drainage is inserted at the time of operation or just after an operation. In this patient, the CSF drainage line was inserted 24 hours before systemic heparinization; therefore, bleeding was probably discontinued before the operation and the extent of the SDH

was minimal, so that recovery was possible without invasive treatment. In our case, headache was the only neurological symptom. Moderately small amounts of subdural blood did not result in neurologic deficit, and headache is a well-known result of procedures that reduce spinal fluid volume and pressure.¹⁰

In our patient, the CSF drainage volume after 24 hours was only 125 mL; however, SDH may have resulted from the CSF drainage. Frequent neurological checks are imperative after CSF drainage catheter insertion. When there is evidence of intracranial hemorrhage in the presence of continuing CSF drainage, CSF drainage should be stopped immediately. Despite these precautions, beneficial effects of CSF drainage have been reported. Less is known about the risks of CSF drainage; thus, CSF drain placement should occur 24 hours before systemic heparinization, and management should be exercised with care, diligence, and caution.

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