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*Case Report*

# Putaminal hemorrhage potentially attributable to over-drainage of cerebrospinal fluid following ventriculoperitoneal shunt surgery in moyamoya disease: A case report

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## ABSTRACT

**Background:** Rebleeding is a prognostic factor in hemorrhagic moyamoya disease (MMD). This report describes a case of cerebrospinal fluid over-drainage from a ventriculoperitoneal (VP) shunt after an intraventricular hemorrhage that may have contributed to a putaminal hemorrhage.

**Case Description:** The patient was a 51-year-old woman with a prior intraventricular hemorrhage and moyamoya diagnosis who had not undergone revascularization surgery and was neurologically stable. She was readmitted in August 2023 with recurrent intraventricular hemorrhage and underwent shunt placement for hydrocephalus. Three weeks later, she developed a massive putaminal hemorrhage and required an emergency craniotomy. The narrowing of the ventricles before the hemorrhage suggested over-drainage. Rapid reduction in intraventricular pressure may have collapsed the fragile collateral vessels and led to the hemorrhage.

**Conclusion:** VP shunt placement for hydrocephalus in patients with MMD following intraventricular hemorrhage necessitates careful management of shunt pressure settings. Given the fragile condition of collateral vessels in MMD, aggressive reductions in shunt pressure should be avoided. Instead, gradual and controlled adjustments are recommended to minimize the risk of over-drainage and subsequent hemorrhagic complications. Regular imaging, such as frequent computed tomography scans, is essential to monitor ventricular size and guide cautious pressure modulation.

**Keywords:** Moyamoya disease, Over-drainage, Putaminal hemorrhage, Ventriculoperitoneal shunt

## INTRODUCTION

Rebleeding is an adverse prognostic factor for patients with hemorrhagic moyamoya disease (MMD). Hemorrhagic MMD has a 7.09% annual rebleeding rate and a 30% rate for a 5-year rebleeding risk.[8]

The Japan Adult Moyamoya (JAM) Trial, a randomized controlled trial conducted in Japan, demonstrated that direct bypass is effective for preventing rebleeding in patients with hemorrhagic MMD.[11] It also classified bleeding sites into anterior and posterior and reported that the rebleeding risk was 8 times higher in the posterior bleeding group than in the anterior bleeding group.[13] An additional analysis by the JAM study group classified and defined three types of angiographic findings (lenticulostriate anastomosis, thalamic anastomosis, and choroidal anastomosis) for abnormally vulnerable collateral tracts as potential bleeding sources. Among these, choroidal anastomosis, which is the most posterior, is the most vulnerable type and prone to rupture, resulting in a high rebleeding rate in the posterior.[3]

In this report, we described a patient with MMD who had two intraventricular hemorrhages due to a ruptured microaneurysm in the peripheral portion of the left anterior choroidal artery and developed a putaminal hemorrhage after ventriculoperitoneal (VP) shunt placement.

## CASE REPORT

### Course before transfer

The patient was a female in her 50s with no significant medical history. Several years ago, she underwent ventricular drainage surgery at another hospital for intraventricular hemorrhage and acute hydrocephalus and was discharged home without any neurological sequelae. She had no history of hypertension or antithrombotic medication use. However, imaging findings suggested MMD. Revascularization surgery was not performed, and she was followed up on an outpatient basis.

In August 2023, she experienced a sudden onset of headache and nausea and was urgently transported to the same hospital. Computed tomography (CT) revealed an intraventricular hemorrhage predominantly centered in the left lateral ventricle [Figure 1]. As the hydrocephalus was mild and her level of consciousness was preserved, ventricular drainage was not performed.

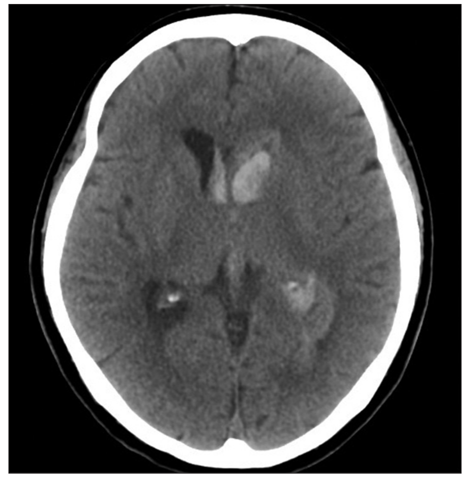

**Figure 1:** Computed tomography image on admission to the previous hospital. The patient experienced intraventricular hemorrhage.

Cerebral angiography indicated bilateral MMD. The anterior and middle cerebral arteries in the left hemisphere were not visible. On the right side, the degree of MMD was milder than on the left side. In addition, the left anterior choroidal artery was well-developed with a peripheral microaneurysm. However, the existence of the lenticulostriate anastomosis could not be clearly identified [Figures 2a and b]. Magnetic resonance angiography (MRA) showed that the cerebral aneurysm was in contact with the left ventricular wall, which possibly caused the intraventricular hemorrhage [Figure 2c]. However, no surgical intervention for the aneurysm was performed.

Although there was no significant increase in intraventricular bleeding during hospitalization, the hydrocephalus gradually worsened and led to drowsiness and cognitive decline. Consequently, a VP shunt was inserted [Figure 3a].

The pressure of the Codman Certas Plus valve (Integra Lifesciences, Plainsboro, NJ, USA) was initially set to 3, and it was adjusted to 2 the next day. Head CT on postoperative day 3 revealed a reduction in the size of the lateral ventricles, although no further adjustments to the valve pressure were made [Figure 3b]. Subsequently, no imaging was conducted for approximately 2 weeks. On postoperative day 19, a head magnetic resonance imaging (MRI) revealed ventricular narrowing, raising suspicion about cerebrospinal fluid (CSF) over-drainage [Figure 3c]. Two days later, the patient suddenly developed right-sided hemiplegia and aphasia. MRI revealed a left putaminal hemorrhage, which led to an urgent referral to our hospital. She had no history of hypertension, and her blood pressure remained well-controlled at a low level following the VP shunt placement. In addition, she was not receiving any antithrombotic therapy, and there were no indications of coagulation abnormalities.

### Course after admission to our hospital

On arrival at our hospital, the patient exhibited severe consciousness disorder, bilateral dilated pupils, complete paralysis of the right upper and lower limbs, and severe aphasia. Head CT revealed a massive putaminal hemorrhage of approximately 100 mL [Figure 4]. For life-saving purposes, an emergency craniotomy was performed for hematoma evacuation.

### Operation

Under general anesthesia, a left frontotemporal craniotomy was performed. The location of the hematoma was identified using intraoperative ultrasound from the cortical surface.

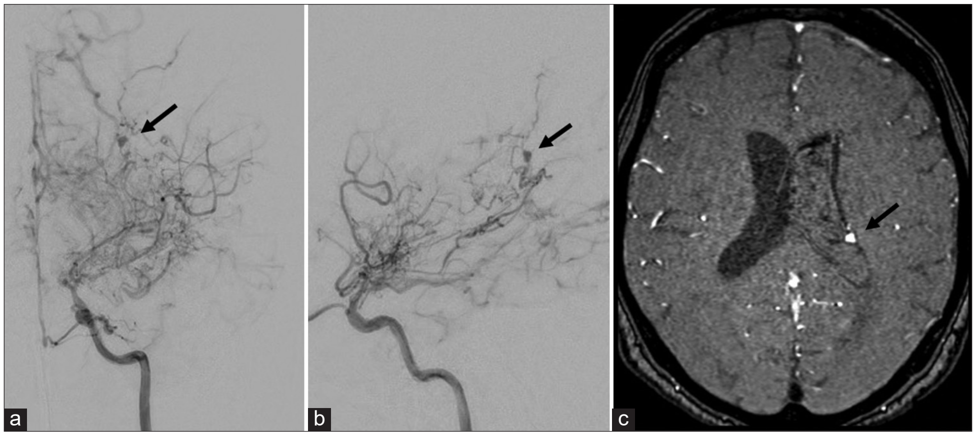

**Figure 2:** Previous hospital images. Digital subtraction angiography (a) frontal view, (b) lateral view. The left internal carotid angiogram shows moyamoya disease and microaneurysm (arrow). (c) Magnetic resonance angiography showing microaneurysm (arrow) in a peripheral anterior choroidal artery within the left lateral ventricle.

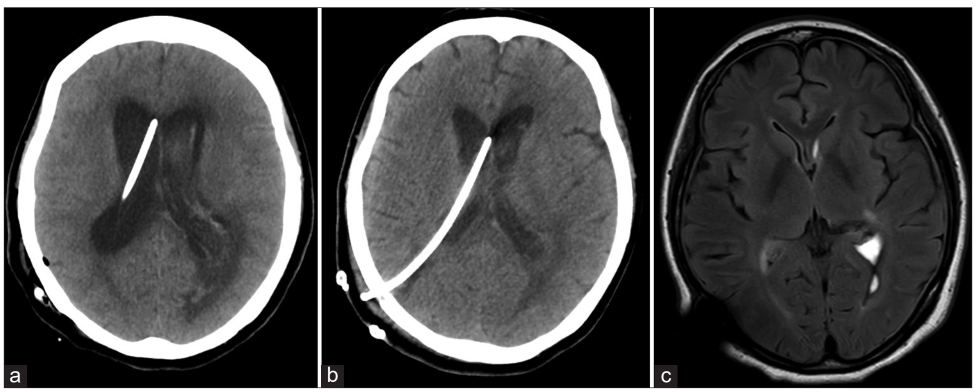

**Figure 3:** After ventriculoperitoneal shunting. (a) Computed tomography (CT) day after ventriculoperitoneal shunt placement; lateral ventricles remain enlarged. (b) CT postoperative day 3, demonstrating size reduction in lateral ventricles. (c) Magnetic resonance imaging 2 days before putaminal hemorrhage onset; ventricle narrowing is observed.

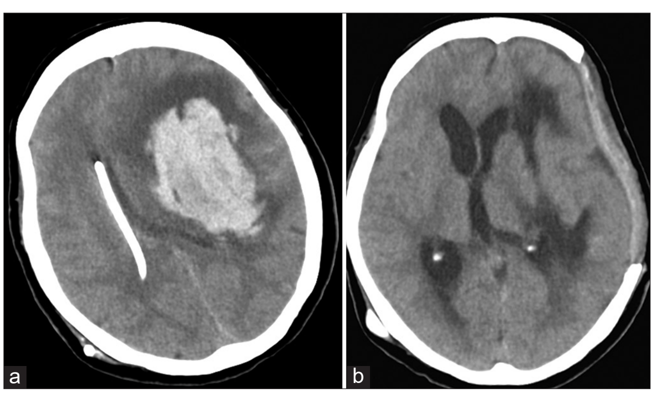

**Figure 4:** Plain computed tomography (CT) head scan after admission at our hospital. (a) CT image at admission showing a giant left putaminal hemorrhage. (b) CT image the day after placement following hematoma evacuation and external decompression.

A cortical incision was made to access the hematoma. As the hematoma was aspirated and removed, a ruptured artery was identified in the deep portion of the hematoma cavity, which was actively bleeding. Hemostasis was achieved by coagulating the artery at its proximal site followed by vessel excision of the vessel. The suspected bleeding vessel was submitted for pathological examination, which revealed no evidence of a microaneurysm or other abnormalities.

The hematoma was generally firm, which presented some difficulty during aspiration and removal. However, achieving hemostasis was relatively straightforward.

### Postoperative course

Postoperative CT revealed that the hematoma had been removed and adequately decompressed. The patient's pupillary findings improved; however, her loss of consciousness did not significantly improve. Tracheostomy was performed 6 days postoperatively, and cranioplasty was performed 23 days postoperatively. During hospitalization, the shunt pressure was adjusted, and the final pressure was set at CERTAS setting 7. Cerebral angiography performed on postoperative day 35 demonstrated that the microaneurysm in the peripheral anterior choroidal artery, which had been identified at the previous hospital, had disappeared.

The sequelae of impaired consciousness, paralysis, and aphasia persisted, and the patient was transferred to the hospital for further rehabilitation on postoperative day 52.

Consent was obtained from the family members of the patient for publication of this case report.

## DISCUSSION

Intracranial ischemia and hemorrhage are the two main manifestations of MMD. Although intracranial hemorrhage is less common than ischemic stroke, it is a major cause of death in patients with MMD.

In this case, the cause of the two episodes of intraventricular hemorrhage was considered to be a microaneurysm rupture in the choroidal anastomosis, which MRA and digital subtraction angiography had already identified. The third cerebral hemorrhage was a putaminal hemorrhage, and its cause was considered to differ from that of the two intraventricular hemorrhages.

There are many reports of repeated bleeding at the same site;[1,6,7] however, there are few case reports of rebleeding at different sites.

In the present case, the time between the second intraventricular and putaminal hemorrhages was approximately 40 days; thus, change in the MMD was unlikely. Intraoperative findings and the pathology of the putaminal hemorrhage showed no microaneurysm in the area assumed to have caused the hemorrhage. VP shunt placement for secondary hydrocephalus was performed 3 weeks before the onset of putaminal hemorrhage, which was suspected to be related.

One complication of VP shunt placement is delayed intracerebral hemorrhage (DICH). DICH is a rare but serious complication following VP shunt insertion with an incidence rate reported between 1.59% and 3.76%.[4,5] DICH typically occurs within 5–13 days postoperatively.[5,10] Identified risk factors include ventricular puncture, catheter malposition, advanced age (≥60 years), and a history of craniotomy.[2,4,5,14] The etiology of early hemorrhage (within 2 days post-surgery) is commonly attributed to venous occlusion caused by intraoperative manipulation, whereas delayed hemorrhage (5–13 days) is more likely associated with brain tissue fragility resulting from underlying brain pathologies.[10]

In the present case, the puncture was performed through the right posterior horn, and it was unlikely related to the cause of the left putaminal hemorrhage. In addition, other potential causes of bleeding, such as hypertension and coagulation abnormalities, were considered unlikely.

Several studies have identified rapid decreases in CSF pressure and brain tissue fragility as key factors contributing to DICH. The pressure exerted by CSF on intracranial blood vessels provides a stabilizing force known as the tamponade effect. A rapid reduction in CSF pressure disrupts this tamponade effect, releasing the compressive support on blood vessels making them more susceptible to rupture and bleeding.[5,9] Particularly, the rapid decrease in CSF pressure following VP shunt valve adjustments may cause fluctuations in intracranial pressure, placing excessive stress on fragile vessel walls and increasing the risk of rupture.[12] Older patients and those with a history of brain disease have increased brain tissue fragility, making them more susceptible to the effects of CSF pressure changes.[5,12]

Although no studies have directly addressed the relationship between MMD and DICH, existing research suggests that the fragile collateral circulation characteristic of MMD may increase the risk of hemorrhage due to the rapid decrease in CSF pressure caused by over-drainage.

A sudden drop in CSF pressure is considered to compromise the normal tamponade effect, releasing the compression on blood vessels and making them more prone to bleeding.[5,9] In particular, after VP shunt valve adjustments the resulting rapid decrease in CSF pressure can lead to intracranial pressure fluctuations, which increase the risk of vessel damage and subsequent hemorrhage.[12] The fragility of brain tissue, especially in patients with a history of traumatic brain injury or prior brain surgeries, is considered another important factor that elevates the risk of hemorrhage.[5] Older patients and those with a history of brain disease have increased brain tissue fragility, making them more susceptible to the effects of CSF pressure changes.[5,12]

Although there have been no reports directly addressing the relationship between MMD and DICH, the fragile collateral circulation observed in MMD is consistent with an increased risk of hemorrhage.

Despite the bilateral narrowing of the lateral ventricles, putaminal hemorrhage occurred in the left hemisphere, where vulnerable collateral vessels were more developed. This is suggestive of an association between CSF over-drainage and the rupture of vulnerable collateral vessels. Therefore, we concluded that over-drainage of CSF excretion due to the VP shunt may have led to disruption of the fragile collateral vasculature in MMD. Routine CT scanning is recommended to prevent DICH after VP shunt placement, and frequent CT follow-ups are considered necessary in patients with MMD who have numerous vulnerable collateral vessels.[2]

In this case, the valve pressure was initially set to a relatively low level and further lowered the next day. Although lateral ventricular shrinkage was observed 3 days after surgery, frequent imaging follow-up was not conducted thereafter. Even in the absence of changes in patient symptoms, regular imaging follow-ups should be performed.

Furthermore, in this case, the shunt valve was not adjusted, even though an MRI performed approximately 2 weeks later showed findings suggestive of CSF over-drainage. The reasons remain unclear. An oversight likely occurred; had the shunt valve pressure been adjusted to a higher setting, the intracerebral hemorrhage that occurred 2 days later might have been prevented. Notably, the presence of lenticulostriate anastomosis is not clear in the image, although it is impossible to deny the possibility that it was involved in the putamen hemorrhage. Nevertheless, VP shunt placement for hydrocephalus in patients with MMD following intraventricular hemorrhage necessitates careful management of shunt pressure settings. Given the fragile condition of collateral vessels in MMD, aggressive reductions in shunt pressure should be avoided. Instead, gradual and controlled adjustments are recommended to minimize the risk of over-drainage and subsequent hemorrhagic complications. Regular imaging, such as frequent CT scans, is essential to monitor ventricular size and guide cautious pressure modulation.

## CONCLUSION

Ventriculoperitoneal (VP) shunting for hydrocephalus in moyamoya disease (MMD) requires careful pressure management to prevent over-drainage and subsequent hemorrhagic complications. In this case, a putaminal hemorrhage occurred following VP shunt placement, with imaging findings suggesting excessive cerebrospinal fluid (CSF) drainage. Although a direct causal relationship remains unclear, rapid CSF pressure reduction may destabilize fragile collateral vessels in MMD. Regular imaging follow-up and gradual pressure adjustments are essential to mitigate these risks. Further studies are needed to establish optimal shunt management strategies in MMD patients at risk of hemorrhagic events.

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