Remote cerebellar hemorrhage after supratentorial craniotomy: illustrative cases

Feng Liu, MS, Dongbo Li, MD, Tao Yang, MD, Congjin Li, BS, Xianhua Luo, MS, Minghui Li, MS, Songlin Wang, MS, Tao Jin, MS, Chunhua Zhang, BS, and Changwen Luo, BS

Department of Neurosurgery, Ankang Central Hospital, Ankang, People's Republic of China

BACKGROUND Remote cerebellar hemorrhage (RCH) is an extremely rare and potentially fatal complication after supratentorial craniotomy. However, the exact pathophysiological mechanism of RCH remains unclear, so clinicians often lack clinical experience in prevention, early diagnosis, and standardized treatment.

OBSERVATIONS The authors retrospectively analyzed data of patients who underwent surgery for supratentorial lesions at their center between 2012 and 2021. They identified 4 patients who developed RCH among 4,075 patients who underwent supratentorial craniotomy. All 4 patients were male, with an average age of 57.5 years. One RCH occurred after tumor resection, and the other 3 occurred after aneurysm clipping. One patient was asymptomatic and received conservative treatment with a favorable outcome. The remaining 3 patients underwent lateral ventricular drainage and/or suboccipital decompression; 2 died, and 1 recovered well.

LESSONS The authors believe that RCH should be considered as a multifactorial cause, and massive cerebrospinal fluid loss plays a key role in the development and progression of RCH. Asymptomatic RCH can be treated conservatively. However, in the case of conscious disturbance, hydrocephalus, and brain stem compression, surgery should be performed immediately. Early detection and individualized treatment would be helpful to avoid potentially fatal outcomes caused by RCH.

Illustrative Cases

Case 1
A 57-year-old male was admitted with a chief complaint of “sudden headache for 2 days.” Cranial computed tomography (CT) revealed a small amount of subarachnoid hemorrhage in the left lateral fissure (Fig. 1A). Emergent digital subtraction angiography (DSA) revealed an aneurysm of the left middle cerebral artery bifurcation, with an aneurysm neck measuring 2.2 mm and height measuring 2.8 mm (Fig. 1B). The patient had no notable medical history and underwent surgical clipping of the aneurysm by left pterional craniotomy without incident. However, postoperative consciousness recovery was delayed. At 8 hours postoperatively, the patient was lethargic with a Glasgow Coma Scale (GCS) score of 11, and a cranial CT examination was performed immediately. On a CT scan, a “zebra-like” hemorrhage was observed in both cerebellar hemispheres (Fig. 1C), accompanied by brain swelling, which was mainly manifested by compression and narrowing of the cisterns around the brainstem. The total amount of drainage fluid in the epidural negative pressure drainage device was about 50 ml within 8 hours after the operation. Conservative treatment was administered under close observation. At 18 hours postoperatively, the

ABBREVIATIONS CSF = cerebrospinal fluid; CT = computed tomography; DSA = digital subtraction angiography; GCS = Glasgow Coma Scale; RCH = remote cerebellar hemorrhage.

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patient's consciousness gradually deteriorated to a shallow coma with a GCS score of 7. A second CT scan showed no increase in intracranial hemorrhage, but hydrocephalus was observed (Fig. 1D). Because the volume of cerebellar hematoma was less than 10 ml and did not increase compared with the previous measurement, it was considered that the decrease in the patient's state of consciousness was caused by hydrocephalus, so lateral ventricular external drainage was decided to be performed immediately. During the operation, about 20 ml of CSF was released, and the pressure was high. However, when the patient returned to the ward after surgery, his condition suddenly deteriorated, accompanied by respiratory arrest, and eventually he died.

**Case 2**

A 47-year-old man underwent endovascular embolization for a ruptured anterior communicating aneurysm 2 years earlier. DSA follow-up revealed a recurrent aneurysm neck with a size of about $3 \times 3$ mm (Fig. 2A). The patient had a history of hypertension for 5 years that was well controlled with medication. The patient underwent left pterional craniotomy to clip the aneurysm without incident (Fig. 2B), and he had stable hypertension throughout the surgery. The patient regained consciousness immediately after surgery, but a routine cranial CT scan 1 hour postoperatively revealed a hematoma in the left cerebellar hemisphere and vermiform region, with compression narrowing the cistern around the brain stem (Fig. 2C). The patient's blood pressure was strictly controlled and treated conservatively. At 8 hours after surgery, the patient presented drowsiness, and his GCS score decreased to 13. An emergent cranial CT examination showed an increase in the left cerebellar hematoma, about 9 ml (Fig. 2D), and posterior fossa decompression was performed immediately. There was no significant change in consciousness after posterior fossa decompression. We noted that the total amount of drainage fluid in the epidural negative pressure drainage device was about 30 ml within 8 hours after aneurysm clipping. However, 27 hours after posterior fossa decompression, the patient's consciousness gradually deteriorated to a shallow coma with a GCS score of 8. A CT scan showed no other abnormalities, but hydrocephalus was observed. Lateral ventricular external drainage was immediately performed, and the patient was continuously administered sedation and analgesia after surgery. Unfortunately, 12 hours later, the patient's condition deteriorated again, with bilateral dilated pupils, accompanied by respiratory arrest, and eventually death. After operation, the lateral ventricular external drainage tube remained unobstructed, and the volume of CSF was about 150 ml.

**Case 3**

A 57-year-old male was diagnosed with an unruptured anterior communicating aneurysm (Fig. 3A), and the finding of his physical examination was negative. Preoperative examination showed no contraindications. The patient underwent surgical clipping of the aneurysm with right pterional craniotomy. No accident occurred during the operation. At 12 hours after surgery, the patient was clearly conscious, and computed tomography angiography examination showed that the aneurysm was clip-closed completely (Fig. 3B), but a cranial CT scan revealed a small amount of hematoma in both the left cerebellar hemisphere and vermis (Fig. 3C and D), without surgical evidence, and conservative treatment was administered under close observation. We observed that the total amount of drainage in the epidural negative pressure drainage device was 90 ml within 12 hours after the operation, and then the drainage flow was strictly
controlled. However, at 36 hours postoperatively, the patient presented with disturbance of consciousness with a GCS score of 10. An emergent cranial CT scan revealed increased hemorrhage in the left cerebellar hemisphere and vermis. At 36 hours postoperatively, CT scan showed increased hematoma in the left cerebellar hemisphere and vermis. CT scan 10 days after posterior fossa decompression showed improvement in brain swelling. Cranial CT follow-up at 1 month postoperatively revealed hydrocephalus with interstitial cerebral edema. Cranial CT follow-up revealed improved hydrocephalus and interstitial cerebral edema after ventriculoperitoneal shunt surgery.

Case 4
A 69-year-old man was admitted with intermittent headache for 3 years. His physical examination showed no significant deficits. Enhanced magnetic resonance imaging of the brain revealed a right sphenoid ridge meningioma (Fig. 4A). The patient had no notable medical history and underwent surgical removal of the tumor with right pterional craniotomy. The patient had stable hemodynamics throughout the surgery. Postoperatively, the patient had no neurological deficits except for mild headache. However, at 18 hours postoperatively, routine cranial CT revealed left cerebellar parenchymal hemorrhage with perihematomal cerebral edema (Fig. 4B and C). At this point, the total amount of drainage fluid in the epidural drainage device was observed to be about 250 ml, and then the epidural drainage tube was closed. Follow-up CT showed no increase in hematoma and no significant mass effect, and the patient was treated conservatively (Fig. 4D). On postoperative day 14, the patient was discharged without neurological deficits. During the 3-month follow-up period, the patient has remained asymptomatic.

Discussion
RCH after supratentorial craniotomy is extremely rare, with a reported incidence of 0.08%–0.6%,1,2 but it is potentially fatal. Moreover, the exact pathophysiological mechanism of RCH remains unclear, so clinicians often lack clinical experience in prevention, early diagnosis, and standardized treatment. Therefore, to improve the success rate of treatment of RCH and reduce the rate of death and disability of RCH, it is necessary to analyze and summarize the experience of RCH-related cases; explore the pathogenesis of RCH; and promote the prevention, diagnosis, and treatment of RCH. Supratentorial craniotomy related to RCH has been reported, including aneurysm clipping,3,4 tumor resection,5,6 hydrocephalus surgery,7 and transcranial pituitary surgery. In our cases, RCH in case 4 occurred after meningioma resection, and the other 3 cases all occurred after aneurysm clipping. The details of all the patients are provided in Table 1.

RCH has no characteristic clinical manifestations, and the diagnosis of RCH depends mainly on cranial CT examination. Patients with a small amount of hemorrhage may be asymptomatic or accompanied by atypical symptoms, such as headache, epilepsy, dyskinesia, and cerebellar signs. The diagnosis of asymptomatic RCH is often found by routine cranial CT scan after surgery. Patients with a large amount of hemorrhage or hydrocephalus present primarily with postoperative changes in consciousness, including delayed awakening from anesthesia, partial restoration of consciousness, persistent coma, or worsening of consciousness after resuscitation from anesthesia. Among our cases, RCH was found in case 1 by cranial CT examination due to delayed awakening from anesthesia, whereas RCH was found for the first time in the other 3 cases without special clinical manifestations by routine postoperative CT examination. Therefore, cranial CT examination should be performed as soon as possible to exclude RCH in patients with delayed anesthesia recovery.
or postoperative deterioration of consciousness. RCH mainly occurs in the early postoperative stage. A retrospective study showed that 46% of patients with RCH developed symptoms within 10 hours after surgery, whereas in 17% of cases, onset may be delayed more than 40 hours after surgery. The most common bleeding pattern for RCH is subarachnoid hemorrhage along the cerebellar sulcus, known as the “zebra sign” because of its striated density of black and white on CT, which is characteristic of RCH. RCH can occur in either unilateral or bilateral cerebellar hemispheres, and it has been reported that bilateral is more common than unilateral, accounting for 55% of cases. In addition, 2 less common types of RCH can be seen on CT scans, including pure cerebellar parenchymal hemorrhage and mixed hemorrhage. A systematic review showed that zebra sign pattern accounted for 64.9%, pure cerebellar parenchymal hemorrhage accounted for 18.5%, and mixed pattern accounted for 16.4%. Among our cases, case 4 presented with pure cerebellar parenchymal hemorrhage, whereas the remaining cases presented with the typical zebra sign.

The exact pathophysiological mechanism giving rise to RCH is unclear. Due to the different forms of RCH hemorrhage, there may be different pathophysiological mechanisms and even multiple factors involved. Currently, it is widely accepted that RCH originates

| Case 1 | Case 2 | Case 3 | Case 4 |
|--------|--------|--------|--------|
| Age, yr/sex | 57/M | 47/M | 57/M | 69/M |
| Hypertension | No | Yes | No | No |
| Factors impairing coagulation | No | No | No | No |
| Anticoagulants/antiplatelet drugs | No | No | No | No |
| Initial diagnosis | Ruptured left MCA aneurysm | Unruptured recurrent ACoA aneurysm | Unruptured ACoA aneurysm | Right sphenoid ridge meningioma |
| CSF management in surgery (EVD/LD) | No | No | No | No |
| Epidural negative pressure drainage device | Yes | Yes | Yes | Yes |
| Volume of drainage before RCH, ml | 50 | 15 | 90 | 250 |
| Symptoms | Delayed recovery from anesthesia | None | None | None |
| Location of RCH | Bilateral hemisphere | Left hemisphere and vermis | Left hemisphere and vermis | Left hemisphere |
| Characteristics of RCH | Zebra | Zebra | Zebra | ICH |
| Timing of bleed, hr | 8 | 1 | 12 | 18 |
| Management | EVD | EVD + SD | EVD + SD | Conservative |
| Outcome (GOS score at discharge) | 1 | 1 | 5 | 5 |

ACoA = anterior communicating artery; EVD = external ventricular drainage; GOS = Glasgow Outcome Scale; ICH = intracerebellar hemorrhage; LD = lumbar drainage; MCA = middle cerebral artery; SD = suboccipital decompression.
from venous hemorrhage. This is due to the cerebellar collapse caused by massive loss of CSF during surgery, which leads to stretching and tearing of the bridging veins that travel along the cerebellar fissure and enter the cerebellar parenchyma. This theory can be used to explain the pathogenesis of the zebra sign bleeding pattern. On a CT scan, the zebra sign is mainly characterized by subarachnoid hemorrhage distributed along the cerebellar cleft with cerebellar edema, which may be caused by venous blood drainage disorder after venous laceration. Moreover, the hemorrhage is mainly distributed in the cerebellar cortex near the tentorium, which is consistent with the distribution of the bridging vein.

However, a pure cerebellar parenchymal hemorrhage may not share the same pathogenesis as a zebra sign pattern hemorrhage. This is because the hemorrhage of the bridging vein stretching tear caused by cerebellar collapse after loss of CSF is a subarachnoid hemorrhage distributed on the cerebellar surface rather than a pure cerebellar parenchymal hemorrhage. In addition, the zebra sign type of RCH is usually bilateral, diffusely distributed in the superior surface sulcus of the cerebellar hemisphere, whereas the pure cerebellar parenchymal type of RCH is unilateral and localized. Because the pure cerebellar parenchymal type of RCH is rarer than the zebra sign type, there currently is no special article to discuss its pathogenesis. A patient with postoperative pituitary adenoma presented with a pure cerebellar parenchyma hemorrhage 12 hours after placement of the lumbar drain, which was considered to be caused by CSF leakage.\(^6\) Pure cerebellar parenchymal hemorrhage was also seen after drilling of a chronic subdural hematoma and was considered to be caused by CSF drainage and intracranial pressure changes.\(^12\) Therefore, it can be inferred that CSF leakage may also be an important cause of the pure cerebellar parenchymal type of RCH. We believe that cerebellar collapse and decreased intracranial pressure caused by massive loss of CSF, resulting in cerebellar ischemic infarction and subsequent hemorrhage transformation, may be the cause of the pure cerebellar parenchymal type of RCH.\(^2,13,14\) In such cases, the location of the hemorrhage is more likely to be the cerebellar parenchyma rather than the subarachnoid space. The pure cerebellar parenchymal type of RCH may also be caused by incidental factors, such as a sudden increase in blood pressure leading to cerebellar parenchymal hemorrhage. There are less common mechanisms that may explain this type of RCH, including coagulopathy, hidden arteriovenous malformation bleeding, and old cerebellar parenchymal injury. In addition, mixed RCH with zebra sign hemorrhage and pure cerebellar parenchymal hemorrhage is also observed on CT scans. So, it seems reasonable that RCH should be considered as a multifactorial cause, and massive CSF loss plays a key role in the development and progression of RCH.

**Observations**

Although most authors agree that rapid and massive CSF loss is the root cause of RCH, they do not describe the details of CSF drainage. The 4 cases we reported had common characteristics: They had no or only a small amount of subarachnoid hemorrhage on preoperative CT, which was a favorable factor for intraoperative CSF loss. It has been reported in many publications that RCH is more common in patients with unruptured aneurysm clipping than in patients with ruptured aneurysm clipping.\(^4,15,16\) This is because in the absence of subarachnoid hemorrhage, loss of CSF is more likely to occur during surgery. All of our cases underwent craniotomy via a pterional approach during which the lateral fissure cisterna was opened and a large amount of CSF was lost. In addition, bilateral frontal air accumulation and brain tissue collapse were observed on the first postoperative CT scan, suggesting a massive loss of CSF during the operation. In terms of the details of CSF drainage, case 4 had excessive CSF drainage before the discovery of RCH, whereas the other cases were within the normal range. The prognosis of RCH cases is different. Most patients with RCH have a good prognosis. However, the overall mortality rate reported after RCH is not negligible (10%–15%).\(^8\) There may also be cases with poor prognosis that are not reported. Both cases 1 and 2 experienced deterioration after lateral ventricular drainage, leading to their eventual death. The cause was considered to be a sudden change of intracranial pressure after external ventricular drainage. Therefore, the presence of severe high intracranial pressure in the posterior fossa must be assessed before lateral ventricular drainage, and the volume and speed of external ventricular drainage must be strictly controlled.

**Lessons**

We believe that RCH should be considered as a multifactorial cause, and massive CSF loss plays a key role in the development and progression of RCH. Because of the lethality of RCH, the prevention and treatment of RCH must be highly emphasized. We believe that the treatment of RCH should be individualized. Conservative treatment is appropriate for asymptomatic patients without intracranial hypertension and brain stem compression. Once RCH is detected, CSF drainage should be strictly controlled immediately to prevent exacerbation. Because some RCH cases deteriorate rapidly, the patient’s blood pressure and consciousness should be closely monitored, and a dynamic cranial CT scan should be performed. At the same time, endotracheal intubation and emergency surgery should be prepared. Surgical procedures include posterior fossa decompression and lateral ventricular external drainage. We believe that the indications of posterior fossa decompression surgery are hematoma volume >10 ml, disturbance of consciousness, disappearance of the peri-brain stem cistern, and compression of the brainstem. Once these conditions occur, posterior fossa decompression should be performed immediately. If suboccipital decompression is to be performed, simultaneous external ventricular drainage should be considered in order to control intracranial hypertension during the operation and prevent postoperative hydrocephalus. Lateral ventricular drainage should be performed immediately when hydrocephalus occurs. If only external ventricular drainage is to be performed, it is important to assess preoperative intracranial pressure in the posterior fossa and to strictly control the speed and volume of CSF drainage postoperatively. The speed and volume of CSF drainage should be strictly controlled after lateral ventricular drainage. The Chinese Expert Consensus on Cerebrospinal Fluid External Drainage in Neurosurgery released in 2018 suggested that the total amount of external ventricular drainage should be about 200 ml, and the average drainage rate should be <15–20 ml/hr.\(^17\) In our experience, it is safe to control the speed <10 ml/hr in patients with RCH.

Although RCH is unpredictable, we have some suggestions that may prevent RCH: (1) Antiplatelet drugs or anticoagulants should be discontinued before surgery to correct coagulation dysfunction; (2) for patients with a history of hypertension, blood pressure should be controlled normally before surgery, and blood pressure should be maintained stable in the perioperative period; (3) avoid massive loss of CSF during surgery; (4) the dura should be sutured safely to control the speed of external ventricular drainage must be strictly controlled.
watertight to avoid leakage of CSF after surgery; and (5) if the cisterna is opened intraoperatively and the dura is not sutured watertight, epidural negative pressure drainage device should be avoided postoperatively. These measures would be helpful to avoid potentially fatal outcomes.

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Author Contributions
Conception and design: D Li, Liu. Acquisition of data: Liu. Analysis and interpretation of data: Liu, Yang, Jin. Drafting the article: Liu. Critically revising the article: C Li. Reviewed submitted version of manuscript: D Li, Liu, X Luo. Approved the final version of the manuscript on behalf of all authors: D Li. Statistical analysis: C Luo. Administrative/technical/material support: M Li, Wang, Zhang. Study supervision: Wang.

Correspondence
Dongbo Li: Ankang Central Hospital, Ankang, People’s Republic of China. lidongbo1103@sina.com.