Remote Cerebellar Hemorrhage after Supratentorial Burr-Hole Trepanation for Unilateral Chronic Subdural Hematoma: Case Report

Manuel Moser and Gerhard Hildebrandt

Remote cerebellar hemorrhage (RCH) after burr-hole evacuation for chronic subdural hematoma (cSDH) is a rare and uncommon complication of minor supratentorial surgery with very few reports in the literature and an uncertain etiology. We present the case of a 62-year-old male who underwent single burr-hole trepanation for unilateral cSDH, revealing incidental RCH on routine postoperative computed tomography (CT) scan most likely resulting from overdrainage of cerebrospinal fluid (CSF) within the postoperative period. The patient recovered well without further neurological intervention. Intra- and postoperative drainage of large volumes of CSF and the venous origin of the bleeding are accepted factors in the controversial concept of its pathophysiology. Alterations in transtentorial pressure and stretching of superficial cerebellar veins with consequent rupture seem to constitute a useful concept, although details on mechanical or hemodynamic changes still remain unknown. A multifactorial etiology with CSF-overdrainage as the major main factor seems reasonable. Neurosurgeons should be aware of the possibility of RCH even in minor supratentorial procedures such as simple burr-hole trepanation. There is a tendency towards more benign courses, but higher patient age and severity of RCH correlate with a poor outcome. Early diagnosis of RCH and close monitoring reduce unnecessary diagnostic and therapeutic interventions in these patients, probably affecting morbidity and mortality.

Keywords: remote cerebellar hemorrhage, chronic subdural hematoma, zebra sign, burr-hole trepanation

Introduction

Burr-hole evacuation for chronic subdural hematoma (cSDH) is a widely used neurosurgical technique beside craniotomy and twist-drill craniostomy for more selected cases.1,2 Although remote cerebellar hemorrhage (RCH) is a known complication of supratentorial and spinal surgery,3,4 to the best of our knowledge only a few cases have been published in the computed tomography (CT) era describing RCH particularly as a postoperative event following burr-hole evacuation for cSDH.5-12 The authors contribute another report to that list, summarize the available cases of RCH in the treatment of cSDH and review the pathophysiology of RCH.

Department of Neurosurgery, Cantonal Hospital St. Gallen, Switzerland

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Case Report

A 62-year-old male presented with a 3-week history of frontal headache, progressive confusion, and gait abnormality due to a slight left-sided hemiparesis. A non-contrast CT scan of the brain revealed a right-hemispheric cSDH (Fig. 1), but no cerebellar abnormality. Five months prior the patient had suffered a mild head trauma without loss of consciousness, imaging of the brain (native CT scan) at that time being negative for any intracranial bleeding, ischemia, or fracture, but showing symmetrical enlargement of the frontal and temporal external cerebrospinal fluid (CSF) spaces due to slight brain atrophy. The medical history of the patient included alcohol abuse, smoking, and depression, the latter being treated previously with Lorazepam (Temesta®, Pfizer AG, Zürich, Switzerland) and Mirtazapine (Remeron®, MSD Merck Sharp & Dohme AG, Luzern, Switzerland). There was no evidence of arterial hypertension or prior hemorrhagic conditions. Preoperative coagulation parameters (prothrombin time (PT), partial thromboplastin time (PTT), international normalized ratio (INR), plated count) were normal. An extended right frontal burr-hole was placed with the patient in supine position. Hematoma evacuation and gentle irrigation with isotonic saline solution was performed using a 10-French silicon drainage, which was placed in the parietal subdural space and connected to closed system drainage without negative pressure at the level of the patient’s head. No intraoperative complications occurred and the patient showed no neurologic deficit after extubation. The subdural drainage aided 80 ml within the

Fig. 1 Pre-operative axial and coronal non-contrast computed tomography scan showing a right-hemispheric chronic subdural hematoma with subacute minor parts, right ventricular compression, diffuse brain oedema of the right hemisphere, and midline-shift to the left (14 mm) and right uncal herniation.
Fig. 2  Axial and coronal non-contrast computed tomography scan of the brain on the second postoperative day revealed a bihemispheric cerebellar hemorrhage, dominating in the right cerebellar hemisphere and the vermis. There was no evidence of cerebrospinal fluid retention due to fourth ventricle compression.

first five postoperative hours and the clinical course was good with the patient claiming minor headache and a little dizziness, being restricted to bed rest in the supine position. Until the nineteenth post-operative hour the subdural drainage aided up to 500 ml, the patient presenting with normal vital parameters and no change in neurological status. The total amount of drained sanguineous fluid did not exceed 500 ml for the next 24 hours. A routine non-contrast CT scan of the brain on the second postoperative day revealed a remote hemorrhage in the cerebellum without fourth ventricle compression (Fig. 2). The subdural drain was removed and a CT-venography was performed which ruled out venous sinus thrombosis. During intensive observation the patient showed no neurological deterioration, but suffered a depressive episode, which led to a restart of the primordial antidepressants. The patient responded well and was transferred to a rehabilitation clinic 1 week after the operation. At a routine follow up, control non-contrast CT scan of the brain showed good postoperative conditions and in-time absorption of the cerebellar hemorrhage (Fig. 3), the neurological exam being unremarkable.

Discussion

The reported incidence of RCH in adult patients undergoing supratentorial surgery is 0.2–0.8%,13–16 with the exception of temporal lobe resection where the incidence is as high as 12.9%.14 The true incidence of RCH might be higher due to asymptomatic patients who do not undergo routine postoperative imaging in some institutions. This is especially the case in spinal surgery with unintended durotomy where postoperative brain imaging is seldom performed because intracranial lesions are unlikely to be considered.19 RCH is located bilaterally (53.5%) as often as unilaterally (46.5%), typically presenting a streaky venous bleeding pattern mostly located in the upper vermis and foliae of the tentorial cerebellar surface which has been termed “zebra sign,”17,18 in contrast to the deeper located hypertensive intracerebellar hematomas which are arterial in origin and generally found near the dentate nucleus.19 The tentorial surface itself is drained by the superior hemispheric and vermian veins.20

Eleven cases of RCH after burr-hole trepanation for cSDH have been reported, which are summarized in Table 1, including the present case.5–12 Mean age at diagnosis was 66 years (range 43–86 years), male individuals being affected more than twice as much as females. Hematoma location was bilateral in 54.5% and unilateral in 45.5%. All patients had received subdural drains. Nine patients (81.8%) showed neurological deterioration leading to emergency CT scan, of which three needed external ventricular drainage (EVD) due to obstructive hydrocephalus. Only two cases, including our patient, were truly incidental on routine postoperative imaging. None of the fully documented cases required posterior fossa decompression and the majority of patients had a modified Rankin Scale of 0 at discharge. The only reported case who succumbed to the disease had undergone three operations for recurrent cSDH within 12 days and in addition to RCH developed intracerebral hematoma, not being deemed suitable for posterior fossa surgery due to poor medical condition and prognosis.7 A review of RCH after supratentorial and spinal surgery reported that symptoms began within the first 10 hours after surgery for 46% of patients and later than 10 hours for 54%.17 Due to the scarce number of reported cases and their retrospective nature there are no established scales to assess the risk of RCH in neurosurgical patients. Risk factors for RCH have been described to be patient-dependent (e.g., hypertension, coagulopathy,
Table 1 Summary of cases of remote cerebellar hemorrhage following burr-hole trepanation for chronic subdural hematoma

| Report           | Age/Sex | cSDH | Subdural drain used | Estimated total volume drained (ml) | Drain in situ (days) | RCH diagnosed by CT after | Clinical presentation | Treatment | Outcome (mRS) on discharge |
|------------------|---------|------|---------------------|-------------------------------------|----------------------|--------------------------|------------------------|-----------|---------------------------|
| Koller et al. (1999) | 59/M    | bilateral | +                    | ~1150                              | 3                     | 3 days                  | pontoocerebellar symptoms | EVD       | 0                         |
| Hur et al. (2003)  | 86/M    | bilateral | +                    | ~510                                | 4                     | 4 days                  | consciousness deteriorated | n.a.      | n.a.                      |
|                   | 75/M    | bilateral | +                    | ~1600                               | 4                     | 5 days                  | consciousness deteriorated | n.a.      | n.a.                      |
| Vogels et al. (2006) | 49/F    | bilateral | +                    | (~20)                               | n.a.                  | 6 hours                 | right-sided ataxia       | EVD       | 0                         |
|                   | 73/M    | right    | +                    | (~40)                               | n.a.                  | 2 days                  | GCS 9, nausea, vomiting   | EVD       | 4                         |
| Hyam et al. (2007) | 79/M    | recurrent right | +                    | n.a.                               | 3                     | 6 days                  | confusion               | conservative | 6                         |
| Chang et al. (2009) | 53/F    | bilateral | +                    | ~220                                | 3                     | 6 hours                 | headache, dizziness, nausea, vomiting | conservative | 0                         |
| Park et al. (2009) | 74/F    | left    | +                    | <300                                | 5                     | 5 days                  | incidental               | conservative | 0                         |
| Kobayashi et al. (2011) | 73/M    | left    | +                    | n.a.                               | 3 hours               | nausea and dizziness    | conservative             | 0                     |
| Kollatos et al. (2011) | 43/M    | bilateral | +                    | ~300                                | 4                     | 1 hour                  | headache, dizziness, nausea | conservative | 0                         |
| Present case      | 65/M    | right    | +                    | ~500                                | 2                     | 2 days                  | incidental               | conservative | 0                         |

M: male, F: female, cSDH: chronic subdural hematoma, CT: computed tomography, EVD: external ventricular drain, GCS: Glasgow Coma Scale, mRS: modified Rankin Scale, n.a.: not available, RCH: remote cerebellar hemorrhage.

Although the exact pathophysiological mechanisms of RCH remain a subject of debate, there is growing consensus that pathological changes in the intracranial venous system lead to venous hemorrhagic infarction. These changes involve an increase of venous and transmural venular pressure primarily resulting from CSF drainage and/or removal of any intracranial mass which led to preoperative elevated intracranial pressure (ICP), increase of intracranial venous pressure as a result of head positioning, transntentorial pressure gradient within the venous system, tearing of superficial and bridging of cerebellar veins, and consequent hemorrhagic venous infarction due to cerebellar “sagging” of superior bridging veins.

Head extension during surgery can lead to a positional occlusion of the internal jugular vein by the transverse process of the atlas, contributing to intracranial venous hypertension. In one neurovascular series of RCH following pterional craniotomy, it was argued that multiple factors such as head positioning and the resulting venous hypertension in conjunction with abundant CSF loss predispose to cerebellar sagging and venous hemorrhagic infarction.

A review of 52 cases of RCH demonstrated that no single presurgical or surgical factor can reliably predict the occurrence of cerebellar hemorrhage after supratentorial craniotomy. Moreover it could be shown that RCH following supratentorial surgery is not an intraoperative complication but more likely occurs as a postoperative event, suggesting that the complication might be precipitated by postoperative suction drainage, which potentially allows a greater volume of CSF to be lost after skull closure than with surgery alone. Nevertheless, the use of CSF drainage after burr-hole evacuation for cSDH is not only safe and associated with reduced recurrence and mortality at 6 months, but also improves patient outcome. Altogether it seems reasonable that RCH should be considered having a multifactorial etiology, CSF overdrainage intra- or more likely postoperatively being the major main factor in our opinion as it was considered the only identifiable factor in the present case and is by far the most cited risk factor in the literature. A meta-analysis of RCH following spinal or supratentorial surgery revealed a good overall prognosis with no remaining neurological deficits in 32.3%, mild deficits in 22.6%, severe deficits in 9.7%, and death in 14.5%, severity of RCH and higher patient age correlating significantly with a poor outcome. RCH after burr-hole trepanation seems to carry a much better prognosis than after craniotomy, with at least 63.6% (n = 7) of documented cases showing full recovery at discharge.
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Conclusion
RCH as a complication of burr-hole trepanation for cSDH is a very rare condition of a most likely multifactorial etiology and a tendency to more benign clinical courses. The awareness of the possibility of RCH even in minor supratentorial procedures and an early diagnosis considering the bleeding pattern are important in reducing its potential mortality. Close monitoring reduces unnecessary diagnostic and therapeutic interventions in these patients, although external ventricular drainage may be crucial in treating obstructive hydrocephalus. We advocate avoidance of extensive head extension and a slow reduction of supratentorial pressure during surgery for cSDH by controlling fluid outflow over time and not inserting suction immediately after opening of the dura, maybe reducing rapid changes in transtentorial pressure gradient. A meticulous registration of drained CSF volume and careful handling of the drainage system should be obligatory in postoperative care to early suspect overdrainage in correlation with neurological deterioration.

Conflicts of Interest Disclosure
The authors declare that they have no financial or other conflicts of interest in relation to this research and its publication. The authors alone are responsible for the content and writing of the article.

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Corresponding author:
Manuel Moser, MD, Department of Neurosurgery, Cantonal Hospital St. Gallen, 95 Rorschacher Strasse, St. Gallen 9007, Switzerland. moser.manuel@gmx.at