Zebra sign: cerebellar bleeding pattern characteristic of cerebrospinal fluid loss

Case report

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Supratentorial subdural hematomas are a rare but well-known complication following spinal interventions. Less often, spinal or supratentorial interventions cause remote cerebellar hemorrhage (RCH). The exact pathomechanism accounting for RCH remains unclear, but an interventional or postinterventional loss of cerebrospinal fluid (CSF) seems to be involved in almost all cases. Hemorrhage is often characterized by a typical, streaky bleeding pattern due to blood spreading in the cerebellar sulci. Three different cases featuring this bleeding pattern following spinal, supratentorial, and thoracic surgery are presented. Possible pathomechanisms leading to RCH are discussed. Based on data from the underlying cases and the reviewed literature, the authors concluded that this zebra-pattern hemorrhage seems to be typical in a postoperative loss of CSF, which should always be considered on presentation of this bleeding pattern.

KEY WORDS • remote cerebellar hemorrhage • cerebrospinal fluid • bleeding pattern

Intracranial pressure due to a loss of CSF, which results in a caudal shift of brain tissue, thus leading to traction and finally rupture of cortical blood vessels. A less frequent complication of spinal or supratentorial surgery is cerebellar hemorrhage distant from the site of surgery. Intraoperative, or more likely postoperative, loss of CSF has been reported in most of these cases and therefore seems to contribute to the pathophysiology of this complication. Spinal as well as supratentorial loss of CSF can produce a similar streaky bleeding pattern together with blood in the sulci of the tentorial surface of the cerebellum. So far, no common pathomechanism has been found to explain the origins of this bleeding pattern in both disease entities. Because this kind of hemorrhage occurs only after spinal or supratentorial loss of CSF, its appearance must always be considered as an indication of its pathogenesis and should be recognized on presentation. We describe three patients who underwent different surgical procedures and suffered secondary cerebellar hemorrhage characterized by the same unique bleeding pattern. The related pathomechanisms and literature are reviewed and discussed.

Case Reports

Case 1

This 52-year-old woman suffering from olisthesis was admitted to our Department of Orthopedic Surgery. After spinal fusion of L5–S1 by using segmental instrumentation, two closed suction drains were placed subcutaneously. Shortly after extubation the patient experienced strong low-back pain, for which she received analgesic medication. Two hours postoperatively the first drain had drawn 350 ml serous fluid tinged with blood and was replaced with a new one. The second drain was replaced 4 hours postoperatively and likewise contained 250 ml serous fluid tinged with blood and was consistent with CSF. Analgesics were counteracted without the desired effect on the patient’s comatose state. An immediate CT scan demonstrated air in the basal cisterns, hydrocephalus, and little blood in both lateral ventricles and the precallosal cistern. An atypical, streaky bleeding pattern was found in the sulci of both cerebellar hemispheres facing the tentorium (Fig. 1). The bleeding was thought to be located within the sulci as well as the cerebellar cortex. Hemorrhage and edema led to compression of the fourth ventricle. Results of an angiography study revealed no vascular abnormality. Assuming a CSF leak, the suction drains were removed. An external ventricular drain was placed and the patient’s status improved during the following 5 days. Her condition then deteriorated after a hypertensive crisis. A CT scan exhibited fresh blood in the frontal interhemispheric fissure and the right lateral ventricle. The patient died of central respiratory paralysis. Autopsy revealed streaky, 3-mm wide or thinner, bilateral blood clots within the subarachnoid space of the upper cerebellar hemispheres and blood within the cervicothoracic subarachnoid space. Furthermore, the right pericallosal artery was found to be eroded by the ventricular drain and thus was responsible for the frontal bleeding.

Case 2

This 58-year-old woman presented with a large left-sided...
temporal meningioma spreading out to the left sphenoid wing and the frontal and parietal bones. She underwent resection of the meningioma and portions of the affected bone and frontotemporal dura mater. Duraplasty was performed and the resected bone was replaced by Palacos. Two drains without suction were placed subcutaneously. When the patient arrived at our intensive care unit, one drain already contained 190 ml serous blood-tinged fluid and the second drain was empty. Two hours later the second drain contained 220 ml similar fluid, which was assumed to be consistent with CSF. Both tubes were then disconnected. The patient vomited repeatedly, although her vital signs were stable. Twenty hours postoperatively the patient underwent her first standard CT scanning study, the results of which displayed postoperative changes and bilateral blood in the sulci of the upper surface of the cerebellum and cerebellar tissue (Fig. 2), which led to compression of the fourth ventricle and occlusive hydrocephalus due to associated edema. No ventricular drain was placed because compression of the fourth ventricle rapidly improved and hydrocephalus was mild. On discharge the patient displayed mild gait ataxia but no additional sign of cerebellar impairment.

Case 3

This 33-year-old woman with left-sided Pancoast tumor underwent chemo- and radiotherapy, complete resection of the residual tumor, and subsequent insertion of a thoracic suction drain. Her state of consciousness deteriorated quickly 1.5 hours postoperatively. She was reintubated and a CT scan demonstrated compression of the fourth ventricle by a predominantly left-sided intracerebellar hemorrhage with accompanying edema, thus resulting in hydrocephalus. Furthermore, blood was located within the sulci of the upper portion of the cerebellum (Fig. 3A and B). An osteoclastic occipital trephination was performed to reduce pressure within the posterior fossa, and the intracerebellar hemorrhage was partially evacuated. Neuropathological examination showed no malignant cells within the evacuated blood, CSF, and small adjacent pieces of cerebellum. An external ventricular drain was placed, and the patient was extubated 2 days later. On the next day the patient’s state of consciousness deteriorated again. A CT scan displayed air in the internal and external CSF spaces (Fig. 3C and D) and the cervical spinal canal (not shown). A subarachnoid–pleural fistula was suspected and repeated thoracotomy exposed an avulsion of the second left thoracic root, which was sealed. The patient was discharged 1 month later with only mild ataxia.

Discussion

Several authors have stated that RCH is more likely to occur in patients who undergo surgery involving the risky process of draining larger volumes of CSF, such as aneurysm surgery or temporal lobectomy. Nevertheless, RCH has also occurred after spinal interventions with planned or occult opening of the dura. Most authors have focused on its appearance only in relation to spinal or supratentorial surgery. Contradictory pathomechanisms have been proposed as causes for both, although a similar bleeding pattern with blood in the sulci of the upper portions of the cerebellum can appear independent of the surgical site. Drain insertion has been found to increase the risk of RCH. This theory is supported by our first two cases, in which the patients lost greater volumes of CSF within a short period of time.

Most theories on the occurrence of RCH indicate involvement of the venous system, a concept supported by the following facts. First, hemorrhage is located in the upper vermian and cerebellar sulci, where the draining veins of the cerebellum are located. Second, although arterial bleeding would be expected to be unilateral, almost all of these hemorrhages were bilateral. In this context, note that RCH has not been reported following lumbar puncture, which has slower CSF loss. Rapid loss of greater amounts of CSF might be needed for cerebellar hemorrhage to occur. Therefore, the favored basic pathomechanism leading to the curvilinear bleeding pattern in the upper cerebellar sulci after spinal surgery indicates a caudal sag of the cerebellum as a result of intra- or postoperative CSF aspiration. Whether stretching leads to occlusion or rupture of bridging cerebellar veins and causes hemorrhagic venous infarction or direct hemorrhage remains unknown.

Poppen, et al., and Madow similarly stated that RCH...
A striped cerebellar bleeding pattern following supratentorial surgery might result in a blockade of venous drainage because of increased supratentorial pressure from, for example, brain swelling, with subsequent venous stasis and hemorrhage. Konig, et al., asserted that the removal of large supratentorial masses or an acute loss of CSF induces a reduction in intracranial pressure, which again leads to a critical increase in the transmural pressure of the veins or venules, which could account for a hemorrhage. Honegger, et al., proposed that a transtentorial pressure gradient is built up by reduced supratentorial and stable infratentorial pressure so that the upper cerebellar veins are traumatized. Following this train of thought, it is difficult to explain how a spinal loss of CSF produces exactly the same bleeding pattern.

Given that spinal and supratentorial loss of CSF result in a similar streaky subarachnoid bleeding pattern (Figs. 1 and 2), an identical pathomechanism should be assumed. Thus, in reviewing the literature and our own cases, the most probable pathomechanism accounting for both—acute spinal or supratentorial loss of CSF—seems to involve stretching of infratentorial cerebellar bridging veins during an upward or downward cerebellar herniation, thus leading to the zebra sign.

Authors of other, less-discussed theories posit that preexisting coagulopathies, postoperative arterial systemic hypertension, or obstruction of the jugular vein from extreme head rotation might cause cerebellar hemorrhage. These factors seem less important but may aggravate or predispose for RCH. None of our patients had a known preexisting coagulopathy, history of arterial hypertension, or diabetes. In all cases drains were inserted, and RCH was assumed to be related to the acute loss of a large amount of CSF, which would correlate with observations made by others who measured an increased amount of drained fluid within a few hours as well. No direct relationship between the volume of fluid drained and the hemorrhage was found in one patient (Case 3) with the intrathoracic suction drain, although the pneumocephalus and intraoperatively detected radicular avulsion definitely supported our assumption that an acute loss of CSF is the underlying reason for RCH. Pneumocephalus, which is frequently observed in patients with subarachnoid-pleural fistulas finally led to a diagnosis, whereas the initial hemorrhage was not interpreted correctly. Diagnosis might have been made earlier had the treating physician recognized the zebra sign as a warning for the rapid loss of CSF.

The outcome in patients with RCH varies significantly and seems primarily to depend on the extent of bleeding, its intracerebellar component, the underlying disease, the amount of time before action is taken, and, of course, further complications, as occurred in one of our cases (Case 1). In cases with blood only in the sulci of the upper cerebellum (as in Case 2), severe complications or serious permanent cerebellar defects are seldom observed. Nevertheless, the greater the extent of intracerebellar hemorrhage, the greater the risk of acute obstructive hydrocephalus and associated complications. Because the exact pathomechanism leading to RCH is not yet fully understood, however, one can only assume that the kinetics and the extent of CSF loss are factors determining the amount of intracerebellar hemorrhage. Therefore, an increased degree of attentiveness should be devoted to postoperative monitoring in patients who have undergone the already mentioned interventions and with a higher risk for this complication, especially patients with a suction drain.

On presentation of the zebra sign, large amounts of CSF have been drained and bleeding has occurred, and thus drainage should be discontinued immediately to prevent aggravation of complications such as cerebellar herniation. In this case it might be better to branch off the drains instead of removing them, because there is still the risk of aggravating symptoms due to additional loss of CSF through the remaining drainage channel. Infusion of Ringer solution to replace lost CSF in case of RCH might prevent further aggravation. On the other hand, once the loss of liquid has been stopped, rapid redistribution between the spinal and supratentorial compartments may be expected, unless occlusive hydrocephalus develops; infusion of fluids in this case might worsen problems. Early placement of a ventricular drain has been beneficial and has improved outcome.

Immediate reexamination to correct a CSF leak might not be required if disconnecting the drains stops the rapid loss of CSF. Only in cases with persistent loss of liquid (as in Case 3) is reexploration inevitable.

**Conclusions**

Quickly pinpointing the correct diagnosis may prevent
the aggravation of complications associated with RCH. Close postoperative monitoring is essential in patients with suction drains and those who have undergone operations associated with an increased risk of RCH. Neuroradiologists and clinicians must be aware that the described streaky, zebra-pattern hemorrhage seems to be characteristic of acute spinal or supratentorial loss of CSF.

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References


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