Case Reports

Suresh Bishokarma, MS, Mch Neurosurgery Resident

Shikher Shrestha

FCPS (Neurosurgery)

Upendra Prasad Devkota

FRCS (Neurosurgery).

Chief Neurosurgeon National Institute of Neurological and Allied Sciences, Bansbari, Kathmandu, Nepal.

Address for correspondence:

Suresh Bishokarma, MS MCH Neurosurgery Resident Neurosurgery Registrar

National Institute of Neurological and Allied Sciences

Bansbari, Nepal P. O. Box: 3711 Tel: +977-1-4373850 Fax: +977-1-4370779

Email: drsureshbk@gmail.com

Date submitted: 26/7/2018 **Date accepted:** 31/7/2018

Remote Cerebellar Haemorrhage after Surgery for Craniopharyngioma: a case report

Postoperative remote cerebellar haemorrhage occurring distant to the operated site as its name suggests is a rare though recognized entity in neurosurgical practice. It can be potentially devastating. Over 100 cases have been described in various literatures over the years. It is postulated to be due to CSF over drainage, has been the most popular theory behind its occurrence. We report a patient who underwent pterional craniotomy for craniopharyngioma, who deteriorated 12 hours following awakening after the surgery, the cause being remote cerebellar haemorrhage.

Keywords: Craniopharyngioma, Remote cerebellar haemorrhage, CSF drainage, Cerebellar sagging, Zebra sign

aemorrhage distant to the operated site is an extremely rare complication. It can be self-limiting if small in size but can be devastating and potentially fatal depending on its size. Number of factors has been implicated for the causation of remote cerebellar haemorrhage (RCH). The exact pathophysiology however is obscure. We present a case operated for suprasellar pathology resulting to RCH and discuss the various hypotheses for its genesis, which have been discussed in various literatures over the year.

Case Report:

A 40-year-old gentleman was admitted to our hospital with insidious onset, gradually progressive, painless diminution in vision starting involving both eyes, associated with frontal headache and loss of libido for 6 months. His blood pressure and sugar levels were in good control. On examination, he had bitemporal loss of vision with diminished acuity of Jaeger's J3 on left and Jaeger's J16on right eye. Rest of the neurological examinations were within normal range.

Magnetic Resonance Imaging (MRI) scan revealed heterogeneously enhancing suprasellar mass without sellar floor enlargement. There was no evidence of hydrocephalus. Preliminary radiological impression of craniopharyngioma was made (as shown in figure 1). Hormonal assay was normal for pituitary hormones. Preoperative visual charting for field documentation showed bitemporal hemianopia. Hisplatelet count and coagulation profile were normal. The patient was started on Dexamethasone preoperatively.

He was operated through standard pterional craniotomy. Tumor was dissected from prechiasmatic, optico carotid, carotico tentorial and trans lamina terminalis corridors. Operative duration was 4 hours. The patient was woken up immediately post operatively and was transferred to intensive care. There was no visual deterioration. Patient however had acute drop in GCS 12 hours following the surgery and had fixed dilated pupils with preserved gag reflex. Postoperative computed tomography (CT) scan showed bilateral cerebellar haemorrhage (as shown in figure 2). Urgent wide midline posterior fossa craniectomy with foramen magnum decompression and evacuation of

Bishokarma et al

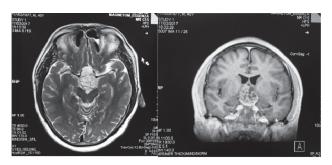




Figure 1: Preoperative MRI scan showing suprasellar lesion, suggestive of craniopharyngioma.

clot was carried out. He was managed in Intensive Care Unit (ICU) with ventilator support for few days but he finally succumbed.

Discussion:

Postoperative hematoma at the operated site is a common occurrence and is related to inadequate haemostasis. Hematoma however, far from the operated site is a rare entity.¹³

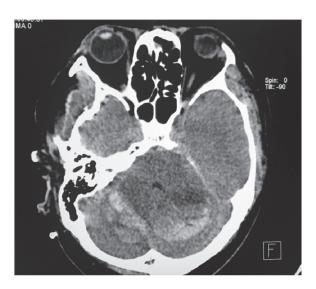


Figure 2: Post-operative scan immediately after deterioration of the patient showing bilateral cerebellar haemorrhage. A typical streaky, curvilinear bleeding pattern with blood in the cerebellar sulci facing the tentorium is seen.

Remote cerebellar haemorrhage (RCH) by definition, is a bleed into the cerebellar parenchyma secondary to neurosurgical intervention at site anatomically unrelated to it.⁵ The reported incidence ranges from 0.08-0.6%.^{7,14} It has been reported in conjunction to various neurosurgical procedures, some of which are aneurysm clipping, temporal lobectomy, spinal surgery, hematoma evacuation, and tumour removal. So far, 4 cases of RCH have been described in association of craniopharyngioma removal in

Author	Age/Sex	Diagnosis	Loss of CSF	Symptom	Treatment	Outcome
Our study	40/M	Craniopharyngioma	+	DOC	Decompressive surgery	Dead
Miyamoto et al.	56/M	Craniopharyngioma	NA	DOC, VI/ IX/X palsy	Decompressive surgery	Good
Konig et al.	42/F	Craniopharyngioma	NA	DOC	EVD	Dead
Tomii et al.	37/M	Craniopharyngioma	+	DOC	Conservative therapy	Good
Friedman et al.	34/M	Craniopharyngioma	NA	DOC	Conservative therapy	Good

Table 1: Summary of 5 cases of remote cerebellar haemorrhage following removal of craniopharyngioma^{6,9,10,15}

various literatures.^{6, 10,9,15}RCH is most frequent in 30-60 years with cases described in as young as 10 years to as old as 83 years.¹

The presentation varies from minor cerebellar signs to decreased level of consciousness and sometimes even prolonged awakening from anaesthesia. Sometimes, it is totally asymptomatic with incidentally detected on postoperative scans. The mortality rate has been reported to be 4.7-7.8%. Figure is slight bilateral preponderance (53.5%) as compared to unilateral occurrence (46.5%). The increased bilateral occurrence of RCH in fact leads to the thinking that the likely pathology for its development is venous. An arterial origin would instead cause unilateral bleeding. Angiography has been performed following postoperative cerebellar haemorrhage and revealed no abnormalities.

The classical pattern - "Zebra sign" distribution of blood along the tentorial surface of cerebellum has also been described by Brockmann et al.³ Most often RCH occurs 10 hours (46%) post operatively.³

The mechanism of RCH has been an enigma. Many postulates have been proposed over the years. Chadduck postulated that sudden rise in blood pressure might cause an increased gradient between the intravascular pressure and CSF pressure leading to haemorrhage into the cerebellar parenchyma.⁴ Andrews and Koci speculated RCH to be due to haemorrhagic conversion of an infarct resulting from transient kinking, spasm or traction of superior cerebellar artery.² Friedman et al in their report of 43 cases proposed that opening of cisterns and the ventricular system causes CSF hypovolemia resulting in cerebellar sagging.¹³ This causes transient occlusion of superior bridging veins of the posterior fossa, leading to subsequent haemorrhagic infarction. Yoshida et al and Konig et al proposed that RCH relates to the amount of CSF drained postoperatively.^{9,16} Postoperative over drainage by negative suctioning may lead to downward displacement of the cerebellum, causing tearing of the superior cerebellar vein and tributaries. Friedman et al suggested preoperative acetylsalicylic acid (ASA) usage and elevated intraoperative systolic blood pressure as possible modifiable risk factors to RCH. While Marquardt et al in their report of 9 patients concluded that there was no relationship between history of hypertension and the occurrence of RCH.¹¹Though direct linear relationship between coagulation parameters and intraoperative hypertensive event have been found, they are hinted to have indirect relationship as for the causation. Considering various proposed mechanism and taking care of them intraoperatively could definitely go a long way in prevention of this potentially fatal complication.

In our case, the patient did not have intraoperative paroxysmal fluctuation of blood pressure or derangement of coagulation parameters. He had received 100 ml of 20% mannitol while opening the dura mater for brain relaxation. Intraoperative CSF drainage from the ventricle was there while opening the lamina terminalis. This probably underscores the importance of gradual decompression to maintain the intracranial pressure within the range.

Conclusion

RCH is a rare but potentially fatal postoperative complication. The precise patho-mechanism is not known. Loss of large volume of CSF especially when the tumour size is large and dehydration due to osmotic diuretics can cause significant brain shift and traction on cerebellar veins to cause infarction and hemorrhagic transformation leading to this rare complication. Timely diagnosis and sensitization of this complication may prevent the dreaded complications as though most smaller hematomas are self-limiting, larger ones might be life threatening.

References

- Amini A, Osborn AG, McCall TD, Couldwell WT. Remote Cerebellar Hemorrhage. American Journal of Neuroradiology. 27:387–390, 2006.
- 2 Andrews RT, Koci TM. Cerebellar herniation and infarction as a complication of an occult postoperative lumbar dural defect. Am J Neuroradiol. 16:1312– 1315, 1995.
- 3 Brockmann MA, Nowak G, Reusche E, Russlies M, Petersen D. Zebrasign: cerebellar bleeding pattern characteristic of cerebrospinal fluid loss. Case report. **J Neurosurg**. 102:1159-62, 2005.
- 4 Chadduck WM. Cerebellar hemorrhage complicating cervical laminectomy. **Neurosurgery**. 9:185–189, 1981.
- Das KK, Nair P, Mehrotra A, Sardhara J, Sahu R N, Jaiswal A K, Kumar R. Remote cerebellar hemorrhage: Report of 2 cases and review of literature. Asian J Neurosurg. 9:161-164, 2014.
- Friedman JA, Piepgras DG, Duke DA, McClelland RL, Bechtle PS, Maher CO et. Al. Remote cerebellar hemorrhage after supratentorial surgery. Neurosurgery. 51 (2):522-523, 2002.
- Honegger J, Zentner J, Spreer J, Carmona H, Schulze-Bonhage A. Cerebellar hemorrhage arising postoperativelyas a complication of supratentorial surgery: a retrospective study. J Neurosurg. 96:248– 254, 2002.

Bishokarma et al

- 8. Koller M, Ortler M, Langmayr J, Twerdy K. Posteriorfossa haemorrhage after supratentorial surgeryreport of three cases and review of the literature. **ActaNeurochir (Wien).** 141: 587 – 592, 1999.
- Konig A, Laas R, Herrmann HD. Cerebellar haemorrhage as acomplication after supratentorial craniotomy. Acta Neurochir. 88:104–108, 1987.
- Miyamoto Y, Nakasu S, Nakasu Y, Handa J. Postoperative intracerebral hematoma remote from the site of craniotomy. Neurologia Medico-Chirurgica. 25 (3):219–222, 1985.
- 11. Marquardt G, Setzer M, Schick U, Seifert V. Cerebellar hemorrhage after supratentorial craniotomy. **Surg Neurol**. 57:241–251, 2002.
- Park JS, Hwang JH, Park J, Hamm IS, Park YM. Remote cerebellar hemorrhage complicated after supratentorialsurgery: retrospective study with

- review of articles. **Journal of Korean Neurosurgical Society**. 46 (2):136–143, 2009.
- 13. Rezazadeh A, Rohani M, Tahamy SA. Remote cerebellar hemorrhage. **Arch Iran Med**. 14:292-3, 2011.
- Toczek MT, Morrell MJ,Silverberg GA,Lowe GM. Cerebellar hemorrhage complicating temporal lobectomy: report of four cases. Journal of Neurosurgery. 85 (4):718–722, 1996.
- Tomii M, Nakajima M, Ikeuchi S, Ogawa T, Abe T. Infratentorial hemorrhage following supratentorial surgery. Neurological Surgery. 27 (10):921–925, 1999.
- Yoshida S, Yonekawa Y, Yamashita K, Ihara I, Morooka Y. Cerebellar hemorrhage after supratentorial craniotomy—report of three cases.
 Neurol Med Chir. 30:738–743, 1990.