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CASE REPORT

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Intraoperative hemorrhage in medulloblastoma: a case report and review of the literature

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Abstract *Object:* We present the first case of intraoperative hemorrhage in a medulloblastoma. *Case report:* A 10-year-old girl presented with a 4-week history of headache, nausea, and vomiting. Radiological examination showed a space-occupying mass in the cerebellar vermis. Surgical removal was performed via a midline suboccipital approach. When the dura was incised and the occipital sinus was ligated after suboccipital craniectomy, bleeding occurred in the tumor. Macroscopically, hematoma was found only in the left part of the tumor and not in the right part. Microscopically, different architectures of tumor vessels, thin-walled and thick-walled, were found between the left part and the right part, respectively. The tumoral contents and hematoma were totally removed. Histological examination revealed a medulloblastoma. *Conclusion:* We experienced a very rare case of medulloblastoma in which intratumoral hemorrhage occurred during operation. We speculate that ligation of the occipital sinus and thin-walled vessels within the tumor might have caused the hemorrhage in our case.

Keywords Intratumoral hemorrhage · Medulloblastoma · Occipital sinus · Midline suboccipital approach · Tumor vessels

Introduction

Intratumoral hemorrhage in medulloblastoma is reported to be extremely rare [15]. Recently, we encountered a case of medulloblastoma, in which a hemorrhage occurred during operation. To our knowledge, intraoperative hemorrhage in medulloblastoma has never been reported. We describe the radiological findings, the operative course and the histological features of this case. We also review the literature on causes of hemorrhage in medulloblastoma and discuss the possible mechanism of hemorrhage in this case.

Case report

A 10-year-old, healthy girl had a 4-week history of headache, nausea and vomiting. Subsequently, she was transferred from a private clinic and immediately admitted to our hospital.

Examination

Neurological examination revealed mild truncal ataxia and papilledema. A computerized tomography (CT) scan showed an isodense, round mass in the cerebellar vermis with obstructive hydrocephalus (Fig. 1a). In addition, inhomogeneous contrast enhancement was revealed (Fig. 1b). A T1-weighted magnetic resonance image showed a hypointense mass in the cerebellar vermis (Fig. 2a). A distinct difference in gadolinium enhancement between the right inferior part and the left superior part of the mass was revealed (Fig. 2b–d). Angiography disclosed tumor stains fed mainly by right anterior and posterior inferior cerebellar arteries and draining toward the bilateral inferior vermian veins and petrosal sinuses. A single occipital sinus was found in the midline of the posterior cranial fossa (Fig. 3).

Operation

First, ventriculostomy was performed to control the intracranial pressure. However, cerebrospinal fluid was not drained, to avoid upward herniation. Then midline suboccipital craniectomy was

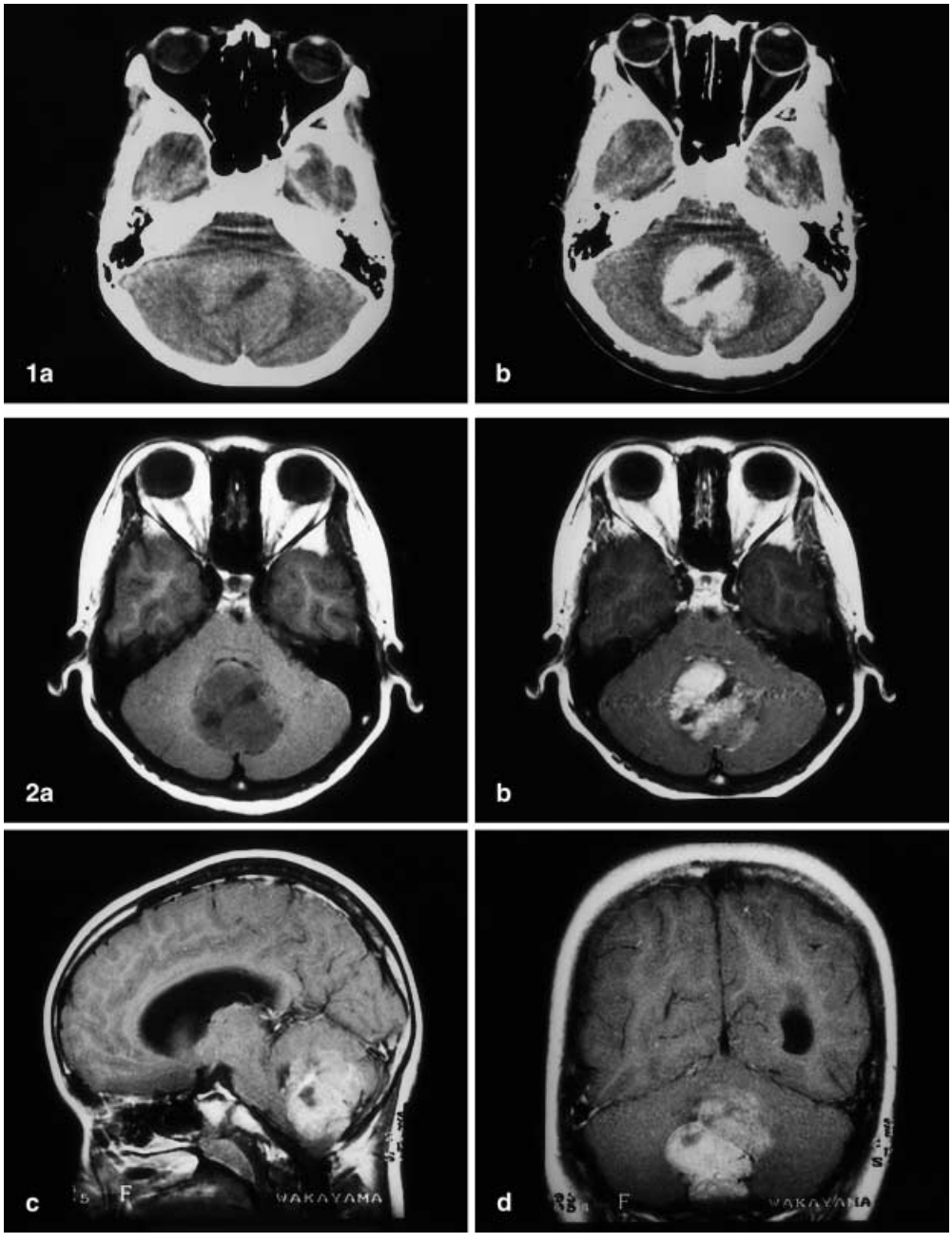

Fig. 1 **a** Plain and **b** enhanced CT scans revealing a round mass in the cerebellar vermis

Fig. 2 **a** T1-weighted MR image and **b–d** gadolinium-enhanced images, revealing a heterogeneously enhanced mass in the cerebellar vermis

carried out. The dura mater was mildly tensioned and was incised with the occipital sinus ligated. Immediately after this procedure, the cerebellum became progressively more tense and bulged especially in the left part (Fig. 4a). Blood-stained cerebrospinal fluid was also drained. Internal decompression was performed by partial excision of the left hemisphere. Intratumoral hematoma was revealed only in the left part of the tumor, and not in the right part. The tumor content and the hematoma were totally removed.

Postoperative course

After operation, only a mild left cerebellar sign was seen. Follow-up CT showed intraventricular hemorrhage (Fig. 4b). Subsequently, chemoradiation therapy was followed, and the girl was discharged without neurological deficits 8 months later.

Histology

Microscopic examination revealed a highly cellular tumor composed of primitive neuroepithelial cells with scant cytoplasm and hyperchromatic nuclei, showing perivascular arrangements. Mitotic figures and apoptotic features were frequently seen. Among these tumor cells there were many blood vessels. In the right part of the tumor, blood vessels showed thick perivascular walls with abundant mesenchymal cells (Fig. 5a). In the left part, thin-walled blood vessels were found, which were associated with massive recent hemorrhages and congestion (Fig. 5b).

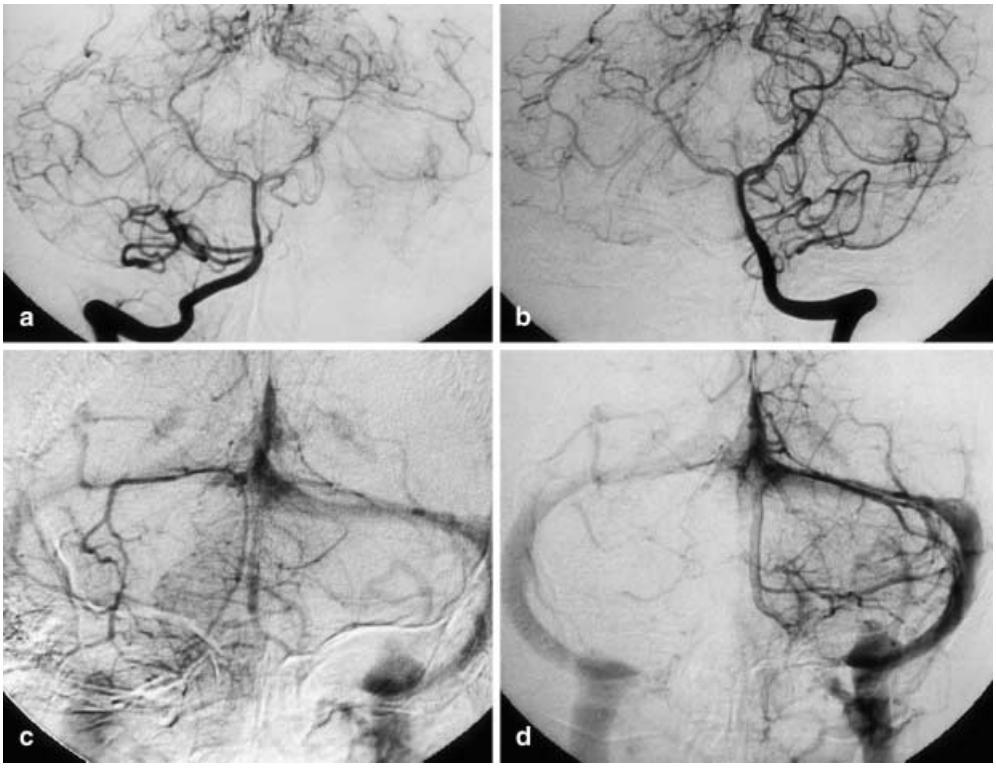

Fig. 3 **a**, **c** Right vertebral arteriogram showing tumor stain fed by right anterior/posterior inferior cerebellar arteries and draining toward bilateral inferior vermian veins and petrosal veins. **b**, **d** Left vertebral arteriogram showing a single occipital sinus in the midline of the posterior cranial fossa

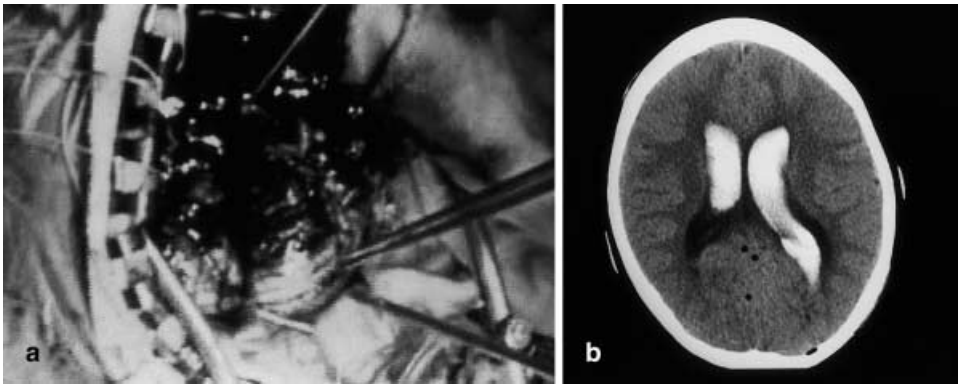

Fig. 4 **a** Intraoperative findings. The cerebellum was bulging progressively from immediately after dural incision and ligation of the occipital sinus. **b** Postoperative CT revealing intraventricular hemorrhage

Discussion

Hemorrhages in brain tumors are reported to occur with a frequency of 2.4–5.1% [4, 18], and intratumoral hemorrhages are considered more likely to occur in rapidly growing malignant or highly vascularized tumors [7, 10, 14]. However, hemorrhages in medullobastomas are rarely found. Zülch stated that hemorrhages do not occur in this tumor [22], and Rubinstein maintained that hemorrhage is uncommon [12]. To our knowledge, only 30 cases of medulloblastomas with hemorrhage have been reported [1, 2, 3, 6, 8, 9, 10, 11, 13, 14, 15, 16, 17, 18, 19, 20, 21], and in 8 cases some authors have discussed the cause and mechanism of hemorrhage (Table 1) [9, 10, 13, 14, 15, 16, 17]. They propose the possibility that hemorrhage might be caused by external ventricular drainage, subtotal removal, or radiation [10, 13, 14, 16]. Spontaneous hemorrhages without any warning signs have been thought to occur as a result of upward herniation, or vascular invasion by tumor cells [9, 15, 18]. Previously, it was reported that a variety of vascular changes can occur within the tumors, resulting in infarction, necrosis, and hemorrhage [21]. These vascular changes, which are less common in medulloblastomas, are: irregular endothelial proliferation obliterating the vascular lumen, thin-walled and often weak vessels subjected to disruption when stretched and distorted by the tumor, and invasion of tumor cells into vessels' walls [3, 13,

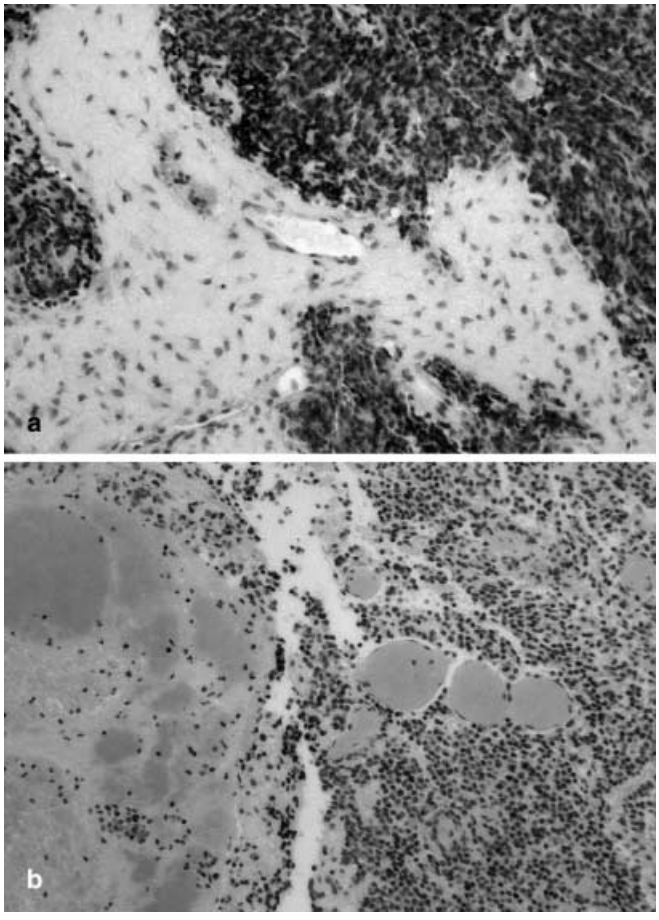

Fig. 5a, b Photomicrographs of the tumors. A highly cellular tumor composed of primitive neuroepithelial cells with scant cytoplasm and hyperchromatic nuclei. **a** The right part of the tumor showing thick-walled vessels with perivascular mesenchymal cells. (H-E, ×200). **b** The left part of the tumor showing thin-walled, fragile vessels with dilatation and massive hemorrhages. (H-E, ×80)

15]. In a few cases of medulloblastoma with hemorrhage, histological investigations have been conducted and the results reported, including secondary changes following operation and/or radiation [13, 14, 15]. In contrast, some reports state that there are no findings to account for a hemorrhage [9, 13].

Our case showed two characteristic features that have never been previously reported. First, the hemorrhage occurred during an operation, apparently being triggered by ligation of the occipital sinus. We can hardly conceive of the hemorrhage as spontaneous. There were no problems with the patient's position and hemodynamics during operation. Surgical procedures, including ventriculostomy before ligation of the occipital sinus, were less likely to be responsible for hemorrhage, considering the unchanged dural tension reflecting ICP. Angioanatomically, the occipital sinus appeared to function as a drain from the tumor. If this is so, ligation of the occipital sinus would disturb the venous drainage and elevate the venous pressure. Secondly, according to radiological appearances, a single tumor had two parts, each of which proved to have a histologically distinct architecture of vessels: hemorrhage occurred in the part of the tumor with thin-walled vessels, and not in the part with thick-walled vessels. The possible mechanism of hemorrhage in our case is disturbance of the venous drainage triggered by ligation of the occipital sinus, inducing rupture of the thin-walled, fragile tumor vessels and thus resulting in intratumoral hemorrhage.

In the midline suboccipital approach to cerebellar tumors, a Y-shaped dural incision of the occipital sinus with ligation of the lower portion running paramedially is usually made [4]. However, in a case with drainage to the well-developed occipital sinus, the possibility of hemorrhage from the tumor should be considered in this operative procedure. Massive hemorrhages in the posterior cranial fossa can be fatal [13, 14]. To avoid this catastrophic complication, dural opening without interruption of the sinus drainage may be preferable. Anatomical variations of the occipital and marginal sinuses must be borne in mind when the dural incision is planned. If the

Table 1 Summary of eight reported cases with hemorrhage in medulloblastoma (*ND* not described)

Reference	Patient age (years)/sex	Cause and mechanism	Histological findings of tumor vessels	Outcome
[10]	9/female	Operation	ND	Died
[17]	6 /male	Spontaneous	Intact vascular walls	Died
[16]	11/female	Ventricular drainage	ND	Died
[14]	9/female	Irradiation	Dilated irregular vessels with thin-walled sinusoids	Died
[9]	11/female	Upward herniation	No abnormal findings	Died
[13]	2/female	Obscure	No abnormal findings	Died
	13/male	Operation and irradiation	Dilated cavernous vessels	Died
[15]	8/female	Vascular invasion by tumor cells	Abnormal vascular components invaded by tumor cells	Died
Present case	10/female	Ligation of the occipital sinus	Thin-walled blood vessels	Alive

necessary exposure is not obtained and dural opening with sinus ligation is required, temporary clips on the sinus will be useful for evaluation of the effects on venous return, to warn of impending edema or hemorrhage.

Ventriculostomy may be necessary to control the intracranial pressure. In our case, as mentioned earlier, ventriculostomy was less likely to be responsible for hemorrhage. However, two cases of intratumoral hemorrhage in posterior fossa tumors a few hours after insertion of an external ventricular drainage tube are documented [16]. Since placement of an intraventicular drain during the posterior fossa procedure carries the risks of upward herniation and intratumoral hemorrhage, although such complications may be prevented by clamping or limiting the cerebrospinal fluid drainage, extreme care should be taken at the time of precraniotomy ventriculostomy in surgical treatment of huge infratentorial mass lesions.

Conclusion

We report a very rare case of intratumoral hemorrhage in medulloblastoma during operation. The hemorrhage is likely to have been caused by ligation of the occipital sinus and rupture of thin-walled, fragile vessels within the tumor.

References

1. Carrieri G, Cazzato G (1960) On a case of hemorrhagic medulloblastoma of the cerebellum. J Psychiatry Neuropathol 88:871–884
2. Chugani HT, Rosemblat AM, Lavenstein BL, Palumbo FM, Luessenhop AJ, Manz HJ (1984) Childhood medulloblastoma presenting with hemorrhage. Childs Brain 11:135–140
3. Glass B, Abbot K (1955) Subarachnoid hemorrhage consequent to intracranial tumors: review of literature and report of seven cases. Arch Neurol Psychiatry 73:369–379
4. Kelly DL Jr, Lyerly MA (1993) Problematical intraoperative events. In: Apuzzo MLJ (eds) Brain surgery. Complication avoidance and management. Churchill Livingstone, New York, pp 1671–1689
5. Kothbauer P, Jellinger K, Flament H (1979) Primary brain tumor presenting as spontaneous intracerebral hemorrhage. Acta Neurochir (Wien) 49:35–45
6. Laurent JP, Bruce DA, Schut L (1981) Hemorrhagic brain tumors in pediatric patients. Childs Brain 8:263–270
7. Little JR, Dial B, Belanger G, Carpenter S (1979) Brain hemorrhage from intracranial tumor. Stroke 10:283–288
8. McCormick W, Ugajin K (1967) Fatal hemorrhage into a medulloblastoma. J Neurosurg 26:78–81
9. Narumi S, Hirano A, Llena JF, Kato T, Yamamoto T (1987) An autopsy case of medulloblastoma with massive hemorrhage. No Shinkei Geka 15:103–107
10. Oldberg E (1933) Hemorrhage into gliomas. A review of eight hundred and thirty-two consecutive verified cases of glioma. Arch Neurol Psychiatry 30:1061–1073
11. Park TS, Hoffman HJ, Hendrick EB, Humphreys RP, Becker L (1983) Medulloblastoma: clinical presentation and management. J Neurosurg 58:543–552
12. Rubinstein LJ (1972) Tumors of the central nervous system. (Atlas of tumor pathology, 2nd ser, fasc 6) Armed Forces Institute of Pathology, Washington, DC, pp 132
13. Shiino A, Nakasu S, Matsuda M, Handa J (1987) Intratumoral hemorrhage in medulloblastoma. Report of two cases. Arch Jpn Chir 56:531–537
14. Sugawara T, Shingai J, Ogawa A, Wada T, Namiki T, Suzuki J (1986) A case of massive fatal hemorrhage in a recurrent medulloblastoma during radiotherapy. No Shinkei Geka 14:109–113
15. Tachibana O, Oki H, Hayashi Y, Nonomura A, Yamashita T, Yamashita J (1990) Repetitive intratumoral hemorrhage in medulloblastoma (case report). Surg Neurol 33:378–383
16. Vaquero J, Cabezudo JM, DeSola RG, Nombela L (1981) Intratumoral hemorrhage in posterior fossa tumors after ventricular drainage. Report of two cases. J Neurosurg 54:406–408
17. Viratchai C, Shuangshoti S (1978) Massive spontaneous hemorrhage in primary intracranial neoplasm. J Med Assoc Thai 61:115–122
18. Wakai S, Yamakawa K, Manaka S, Takakura K (1982) Spontaneous intracranial hemorrhage caused by brain tumor: its incidence and clinical significance. Neurosurgery 10:437–444
19. Weinstein ZR, Downey EF Jr (1983) Spontaneous hemorrhage in medulloblastomas. AJNR Am J Neuroradiol 4:986–988
20. Zee CS, Segall HD, Miller C, Ahmadi J, McComb JG, Han JS, Park SH (1982) Less common CT features of medulloblastoma. Radiology 144:97–102
21. Zimmerman RA, Bilaniuk LT (1980) Computed tomography of acute intratumoral hemorrhage. Radiology 135:355–359
22. Zülch K (1986) Brain tumors. Their biology and pathology, 3rd edn. Springer, Berlin Heidelberg New York, pp 328