

## Successful treatment of a ruptured flow-related aneurysm in a patient with hemangioblastoma: Case report and review of literature

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### Abstract

**Background:** No cerebral aneurysms on the feeder associated with hemangioblastomas that ruptured before resection have been reported. We report a patient with a ruptured flow-related aneurysm associated with cerebellar hemangioblastoma and a tumor feeder treated simultaneously by a single procedure of embolization using *N*-butyl cyanoacrylate before tumor removal.

**Case Description:** A 36-year-old female with a cerebellar tumor was admitted to our institute. Four days later, she suffered a massive subarachnoid hemorrhage mainly in the posterior fossa. Left vertebral angiograms showed an aneurysm on the feeding artery, posterior inferior cerebellar artery. Both the aneurysm and its main feeder were simultaneously treated by a single procedure of embolization using *N*-butyl cyanoacrylate. Their complete obliteration was confirmed angiographically. Four days after the procedure, we removed the tumor and the embolized aneurysm. The pathological diagnosis was hemangioblastoma and flow-related ruptured aneurysm.

**Conclusion:** Cerebral angiography should be performed to rule out vascular abnormalities such as cerebral aneurysms adjacent to the tumor in patients with hemangioblastoma who present with intracranial hemorrhage. We emphasize the usefulness of embolization with *N*-butyl cyanoacrylate for hemangioblastoma with ruptured feeder aneurysm, by which the aneurysm and the feeder could be simultaneously embolized.

**Key Words:** Cerebral aneurysm, embolization, hemangioblastoma, *N*-butyl cyanoacrylate, subarachnoid hemorrhage

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### INTRODUCTION

Cerebral aneurysms associated with hemangioblastomas are very rare<sup>[4,7,8,13,15,16]</sup> and to our knowledge no cerebral

aneurysms on the feeder that ruptured before resection have been reported. We encountered a patient with hemangioblastoma with a ruptured distal posterior inferior cerebellar artery aneurysm on the main feeder.

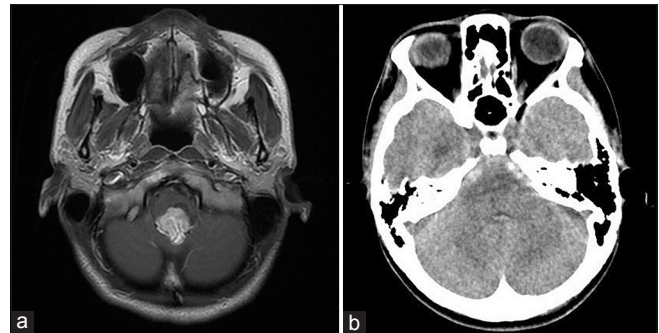
Our treatment consisted of embolization with N-butyl cyanoacrylate and subsequent surgical removal.

## CASE REPORT

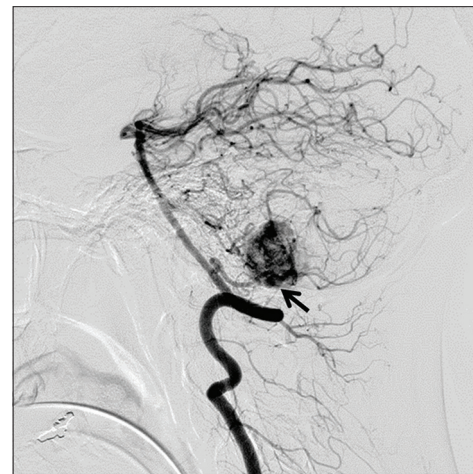
A 36-year-old healthy female was admitted to our hospital due to the sudden onset of a severe headache. Gadolinium-enhanced magnetic resonance imaging (MRI) revealed an enhanced mass in the cerebellar vermis [Figure 1a]. Four days later, her consciousness became impaired and she suffered generalized seizures. Brain computed tomography (CT) showed subarachnoid hemorrhage (SAH) in the posterior fossa [Figure 1b]. We performed cerebral angiography to ascertain the peritumoral angioarchitecture. A left vertebral angiogram showed a tumor stain in the cerebellar vermis fed by the posterior inferior cerebellar artery (PICA) and an aneurysm on the distal portion of the feeder [Figure 2]. We considered a flow-related aneurysm as the origin of the hemorrhage and performed endovascular treatment of both the feeder and the aneurysm using a N-butyl cyanoacrylate lipiodol mixture to avoid rebleeding and to minimize intraoperative blood loss. Under general anesthesia, a 5 Fr Guider™ guiding catheter (Stryker, Fremont, California, USA) was inserted in the left vertebral artery. After positioning a Magic™ micro catheter (BALT, Montmorency, France) in the distal portion of the vermian branch of PICA just proximal to the aneurysm, we carefully injected a 20% NBCA-lipiodol mixture into the aneurysm and the distal portion of the main feeder. Postoperative left vertebral angiograms confirmed the complete disappearance of the aneurysm; the anterior part of the tumor stain was clearly diminished [Figure 3a-d]. Her clinical condition gradually improved and she suffered no recurrent hemorrhage. Four days later, we removed the vascular-rich tumor attached to the cerebellar vermis and the embolized distal PICA aneurysm [Figure 4a]. Pathological findings confirmed our preoperative diagnosis of cerebellar hemangioblastoma and intracerebral aneurysm, and showed disruption of the internal elastic lamina and rupture of the adventitia [Figure 4b].

## DISCUSSION

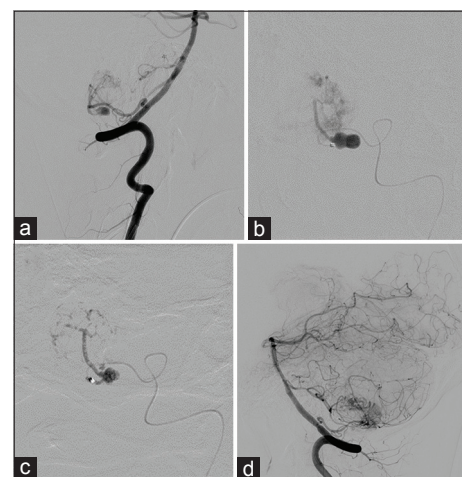
Glacker *et al.*<sup>[3]</sup> reported that the probability of spontaneous hemorrhage from hemangioblastomas is very low (0.024%/person/year) and that lesions smaller than 1.5 cm present no risk for spontaneous hemorrhage. Despite of the report, our patient suffered recurrent hemorrhages despite the small size of her tumor. We posit that flow-related aneurysms associated with hemangioblastomas can be the cause of bleeding even if the tumor is small, therefore we recommend preoperative studies such as cerebral angiography or



**Figure 1:** (a) Gadolinium-enhanced magnetic resonance imaging (MRI) revealed an enhanced mass in the cerebellar vermis. (b) Brain CT image revealed massive SAH in the posterior fossa after rebleeding



**Figure 2:** Preoperative left vertebral angiogram note the tumor stain. The tumor is fed by the vermian branch of the left posterior inferior cerebellar artery. A flow-related aneurysm (arrow) is seen in the arterial phase

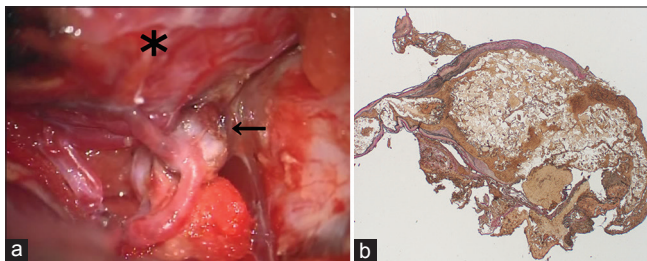


**Figure 3:** Left vertebral angiograms. (a) Intraoperative view before embolization (b) Superselective angiograms from the microcatheter show a dumbbell-shaped aneurysm just distal to the catheter tip. (c) NBCA injection into the aneurysm and the distal and proximal part of the feeder. (d) Complete disappearance of the aneurysm and the anterior part of the tumor stain seen in Figure 2

high resolution enhanced CT to confirm the peritumoral angioarchitecture.

Although many cases about hemorrhage associated with hemangioblastomas have been reported in patients without other vascular anomalies, only six studies<sup>[4,7,8,13,15,16]</sup> documented aneurysms associated with hemangioblastomas. Only two patients with rupture suffered aneurysmal SAH 5 years<sup>[7]</sup> and 23 months<sup>[15]</sup> after tumor removal [Table 1]. De San Pedro *et al.*<sup>[2]</sup> concluded from the literature that aneurysms associated with hemangioblastomas were a negligible source of hemorrhage. In our patient, aneurysmal wall rupture was confirmed pathologically; consequently we thought that the origin of hemorrhage was a ruptured flow-related PICA aneurysm on the tonsillar segment. This is the first case of the rupture, before tumor removal, of a flow-related aneurysm associated with hemangioblastoma.

Cerebral angiographs of intracranial hemangioblastomas typically show intense tumor blush from the feeding arteries and enlargement of the drainers in the late arterial- or capillary phase. These findings are indicative of high tumor vascularity and an increase in the regional blood flow from a slightly enlarged feeding artery to the draining vein. This results in hemodynamic stress that may lead to the formation of an aneurysm on the feeder as like brain arteriovenous



**Figure 4: (a) Intraoperative findings. Note the ruptured aneurysm (arrow) on the feeder near the vascular-rich tumor (asterisk). (b) Microphotograph of the aneurysm showing disruption of the internal elastic lamina and rupture of the adventitia (Elastica van Giesson staining, original magnification ×10)**

malformations (AVMs). In previous large studies of brain AVMs, the incidence of flow-related aneurysms associated with AVMs ranged from 10.9% to 30.7%, and from 45% to 83.3% in patients who manifested hemorrhage.<sup>[5,6,10]</sup> Westphal *et al.*<sup>[14]</sup> and Schmidt *et al.*<sup>[12]</sup> suggested that infratentorial location of flow-related aneurysms associated with AVMs have significantly increased the risk of hemorrhage. As our case, aneurysms associated with hemangioblastomas in the posterior fossa may have higher frequent risk of rupture compared with them in other locations.

Whether flow-related aneurysms are at increased risk for rupture and whether they should be addressed before the treatment of AVMs remains controversial.<sup>[5,6,10]</sup> We suggest that ruptured aneurysms associated with hemangioblastoma should be treated first by embolization or direct surgery when embolization is difficult.

The preoperative embolization of hemangioblastomas has been debated.<sup>[1,9,11]</sup> Cornelius *et al.*<sup>[1]</sup> reported that preoperative embolization of cerebellar hemangioblastomas using particle has a high risk for acute tumor bleeding and death. Sakamoto *et al.*<sup>[11]</sup> reported that the preoperative embolization of hemangioblastomas using glue is superior to embolization with particles. Murai *et al.*<sup>[9]</sup> suggested that endovascular treatment using glue was safe and effective and lowered the risk for post-embolization intratumoral hemorrhage and the recanalization of occluded vessels. In our case, we opted for NBCA as embolic material because it can provide simultaneously the embolization of the aneurysm and the tumor to prevent aneurysmal bleeding and intraoperative hemorrhage during tumor resection. We propose our treatment method if there are no normal branches arising at the more distal part of the feeder.

## CONCLUSION

We encountered a patient with hemangioblastoma and a ruptured aneurysm on the distal PICA, the main feeder.

**Table 1: Previously-reported cases of aneurysm associated with hemangioblastoma**

Authors and year	Age (years)/ gender	Hemorrhage	Feeding vessel	Aneurysm location	Aneurysm classification	Tumor location	Clinical onset
Yoshii <i>et al.</i> , 1976	50 F	SAH after tumor removal	Rt PICA	BA bifurcation Lt ICA bifurcation	Unrelated	Rt cerebellum	Tumoral, SAH
Ueno <i>et al.</i> , 1977	50 F	No	Lt PICA	Lt ICA	Unrelated	Lt cerebellum	Tumoral
Guzman <i>et al.</i> , 1999	53 M	No	Lt PICA, Lt AICA	Lt distal AICA	Distal flow-related	Lt cerebellum	Tumoral
Menovsky <i>et al.</i> , 2002	52 F	SAH after tumor removal	Lt AICA	Lt BA-AICA	Proximal flow-related	Lt cerebellum	Tumoral, SAH
Zager <i>et al.</i> , 2002	53 M	No	Rt AICA	Rt distal AICA	Distal flow-related	Rt cerebellum	Tumoral
Murai <i>et al.</i> , 2006	72 M	No	Rt SCA	Rt ICA-PPTA	Unrelated	Vermis	Tumoral
Our case	37 F	SAH before tumor removal	Lt PICA	Lt distal PICA	Distal flow-related	Vermis	IVH, SAH

Unrelated: Aneurysm on artery unrelated to the hemangioblastoma feeder; proximal flow-related: aneurysm on the proximal portion of a major artery located on the ICA or the circle of Willis or on vertebrobasilar arteries feeding the hemangioblastoma, distal flow-related: aneurysm on the distal portion of the artery feeding the hemangioblastoma. AICA: Anterior inferior cerebellar artery, BA: Basilar artery, ICA: Internal carotid artery, Lt: Left, PICA: Posterior inferior cerebellar artery, PPTA: Persistent primitive trigeminal artery, Rt: Right, SAH: Subarachnoid hemorrhage, F: Female, M: Male

The flow-related aneurysm was treated by embolization using NBCA and the hemangioblastoma was subsequently resected totally. Merit of this treatment is that the aneurysm and the tumor feeder are simultaneously embolized by a single procedure. Cerebral angiographs should be obtained to rule out vascular anomalies near the tumor and to avoid unpredictable intraoperative bleeding in patients with hemangioblastoma and hemorrhage.

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