Persistent Primitive Trigeminal Artery Aneurysm Associated With Cerebellar Hemangioblastoma
—Case Report—

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Abstract
A 72-year-old man presented with a cerebellar vermian tumor manifesting as headaches and vertigo. Angiography disclosed a vascular tumor fed by the superior cerebellar artery and an aneurysm of a primitive trigeminal artery. The patient underwent right occipital craniotomy to remove the highly vascular tumor via an occipital transtentorial approach. Association of a cerebral aneurysm with a hemangioblastoma has been reported previously in only five cases. Only three aneurysms were located on the feeding artery. The aneurysm in this case was not on the feeding artery. Simple coincidence might account for this case.

Key words: intracranial aneurysm, persistent primitive trigeminal artery, hemangioblastoma, cerebellum

Introduction
Association of a cerebral aneurysm with a brain tumor is rare. Cerebral aneurysms have been reported in patients with meningiomas, pituitary adenomas, and other brain tumors. Only five cases involving a hemangioblastoma have been reported (Table 1). The patients were aged 50 to 72 years (mean 55 years). The initial clinical symptoms were due to the hemangioblastomas in all cases. Previously, only one patient had initial clinical symptoms due to hemangioblastoma and subsequent clinical symptoms caused by subarachnoid hemorrhage from cerebral aneurysm 23 months later. Angiography revealed aneurysms in the feeding artery of the hemangioblastomas in three cases.

We treated a patient with a persistent primitive trigeminal artery (PPTA) aneurysm associated with a cerebellar hemangioblastoma.

Case Report
A 72-year-old man with headaches and vertigo was admitted to our hospital. Computed tomography revealed a homogeneously enhanced cerebellar vermian mass surrounded by significant edema. Magnetic resonance imaging also demonstrated a homogeneously enhanced mass in the cerebellar vermis, and an area of flow void related to the right intracavernous portion of the right internal carotid artery (Fig. 1). Right vertebral angiography confirmed the presence of a richly vascular tumor 2.5 cm in diameter, supplied mainly by the right superior cerebellar artery (Fig. 2). Right carotid angiography confirmed the presence of a giant aneurysm at the bifurcation of the internal carotid artery and a PPTA that was not supplying the tumor (Fig. 3). The diagnosis was hemangioblastoma. The patient underwent right occipital craniotomy to remove the highly vascular tumor via an occipital transtentorial approach. The histological diagnosis was hemangioblastoma.

The postoperative course was uneventful. Postoperative vertebral angiography disclosed no residual tumor stain or draining veins. Endovascular balloon occlusion test of the internal carotid artery in conjunction with single photon emission computed tomography and clinical neurological evaluation was performed. The patient did not tolerate the balloon occlusion test. We recommended carotid artery ligation with radial artery grafting for the giant aneurysm, but the patient refused further
Table 1  Reported cases of coexisting hemangioblastoma and cerebral aneurysm

<table>
<thead>
<tr>
<th>Author (Year)</th>
<th>Age/Sex</th>
<th>Site of aneurysm</th>
<th>Site of hemangioblastoma</th>
<th>Feeder of hemangioblastoma</th>
<th>Cause of symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yoshii et al. (1976)</td>
<td>50/F</td>
<td>BA bifurcation, lt ICA terminal</td>
<td>rt cerebellar hemisphere</td>
<td>rt PICA</td>
<td>tumor, SAH from aneurysm</td>
</tr>
<tr>
<td>Ueno et al. (1977)</td>
<td>50/F</td>
<td>lt ICA</td>
<td>lt cerebellar hemisphere</td>
<td>lt PICA</td>
<td>tumor</td>
</tr>
<tr>
<td>Guzman and Grady (1999)</td>
<td>53/M</td>
<td>lt distal AICA</td>
<td>lt cerebellar hemisphere</td>
<td>lt AICA and PICA</td>
<td>tumor</td>
</tr>
<tr>
<td>Menovsky et al. (2002)</td>
<td>52/F</td>
<td>lt VA-AICA</td>
<td>lt cerebellar hemisphere</td>
<td>lt AICA</td>
<td>tumor</td>
</tr>
<tr>
<td>Zager et al. (2002)</td>
<td>53/M</td>
<td>rt distal AICA</td>
<td>rt cerebellar hemisphere</td>
<td>rt AICA</td>
<td>tumor</td>
</tr>
<tr>
<td>Present case</td>
<td>72/M</td>
<td>rt ICA-PPTA</td>
<td>cerebellar vermis</td>
<td>rt SCA</td>
<td>tumor</td>
</tr>
</tbody>
</table>


Fig. 1  Axial T₁-weighted magnetic resonance image with contrast medium demonstrating a signal void in the right cavernous sinus and a homogeneously enhanced lesion in the cerebellar vermis.

Fig. 2  Right vertebral angiogram, anteroposterior view, in the early arterial phase showing a tumor stain in the cerebellar vermis, which is predominantly supplied by the right superior cerebellar artery.

treatment. The patient was discharged in good condition.

Discussion

Various mechanisms have been proposed to account for aneurysm formation associated with hemangioblastomas.10,14 Three previous patients had aneurysms arising from the feeding artery of the hemangioblastoma.10,14 Increased hemodynamic stress exerted on an artery is widely believed to contribute to aneurysm formation,5,14,20 so aneurysms associated with hemangioblastomas may be attributed to high regional blood flow to these tumors, similar to aneurysms associated with arteriovenous malformations (AVMs).14 Both AVM and hemangioblastoma increase blood flow toward the lesion, which would predispose to development of an aneurysm.
Aneurysm and Hemangioblastoma

Fig. 3 Right internal carotid angiogram, oblique view, demonstrating the cavernous internal carotid artery aneurysm and persistent primitive trigeminal artery.

of the feeding vessel. However, high flow in feeding vessels cannot explain all occurrences. Hemangioblastomas constitute of 1% to 2% of all intracranial tumors, and are the most vascular true brain neoplasms. One of the three aneurysms of the feeding artery was associated with cerebellar hemangioma. Multifocal genesis was proposed based on the effects of tumor-secreted signaling molecules like vascular endothelial growth factor and vascular permeability factor, which are expressed by hemangioblastomas. Aneurysms of the distal segment of the anterior inferior cerebellar artery are relatively rare. In two other reported cases and in our own, the aneurysm did not arise from the artery feeding the hemangioblastoma. In our case, carotid angiography revealed the aneurysm at the bifurcation of a PPTA which was not supplying the hemangioblastoma. Therefore, simple coincidence might account for this case.

PPTA is an unusual arterial variant with a reported prevalence of approximately 0.2%. PPTA is associated with various vascular abnormalities such as aneurysms, AVM, and compressive cranial neuropathies. Cerebral aneurysms are the most common lesion associated with PPTA. In contrast, intracranial tumor is extremely rare. PPTA has been associated with pituitary adenoma, meningioma, and astrocytoma, but not with hemangioblastoma. The prevalence of associated cerebral aneurysms in patients with PPTA has been reported as 17% to 30%. However, retrospective evaluation of case reports suggested the prevalence of aneurysms in patients with PPTA was no greater than that in the overall population.

Surgical treatment for PPTA aneurysm is difficult, reflected by the confusing array of reports and recommendations, including carotid artery ligation with or without bypass surgery, and intracranial coil placement with stenting. The unruptured aneurysm was asymptomatic in our elderly patient, and he rejected further intervention. Management has relied on follow up.

References


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