Ruptured anterior communicating artery aneurysm encased in a tuberculum sellae meningioma

Case report


Department of Neurosurgery, Saiseikai Utsunomiya Hospital, Tochigi, Japan

A n association of intracranial aneurysm and brain tumor rarely occurs. Although aneurysms have been observed in patients with gliomas, pituitary adenomas, and other rare brain tumors, the coincidence is highest in patients with meningiomas. A number of reports of meningiomas coexistent with intracranial aneurysms have been published. Subarachnoid hemorrhages (SAHs) in these patients were secondary to the rupture of aneurysms with one rare exception. However, SAH caused by the rupture of an embedded aneurysm has never been reported. We report on an elderly patient who experienced SAH from a ruptured anterior communicating artery (ACoA) aneurysm completely encased in a meningioma in the tuberculum sellae.

Case Report

History. This 70-year-old woman presented at the cardiology outpatient clinic in our hospital with sudden onset of a bifrontal pulsative headache and nausea. She experienced no disturbance of consciousness, cranial nerve abnormalities, motor paresis, or sensory impairment. Emergency computerized tomography (CT) scanning disclosed an SAH and she was referred to our department. The patient had a long history of hypertension and had had a uterine corpus cancer, which had been treated by radical hysterectomy and irradiation 15 years before.

Examination. On admission, the patient was neurologically intact except for right hypossia. A CT scan revealed a diffuse SAH and scattered frontal calcifications. Angiographic studies demonstrated an aneurysm arising from the ACoA and a faint patchy stain around the sella, mainly fed by the posterior ethmoidal artery. The bilateral internal carotid artery (ICA) and anterior cerebral artery (ACA) were shifted posteriorly but no arterial narrowing was observed. A CT scan obtained following angiographic studies demonstrated a clearly circumscribed high-density area above the tuberculum sellae. The T1-weighted magnetic resonance (MR) images disclosed a suprasellar high-intensity lesion, which was hypointense on T2-weighted imaging. Gadolinium-enhanced T1-weighted MR images demonstrated that the aneurysm was completely encased in the tumor.

KEY WORDS • aneurysm • anterior communicating artery • meningioma • subarachnoid hemorrhage • tuberculum sellae
Operation. The operation was performed on the 20th day post-SAH. While the patient received a general anesthetic, a coronal skin incision was made followed by a bilateral frontal craniotomy. Via the subfrontal and interhemispheric approaches, a reddish-gray tumor was encountered between the olfactory nerves. After piecemeal resection, the bilateral optic nerves and carotid arteries were identified. Although the tumor encased the bilateral ACAs, the ACoA, and the Heubner arteries, there was no adhesion to any of these vessels. The bilateral $A_1$ was temporarily clipped and the aneurysm, which originated from the left $A_1-A_2$ junction (Fig 4. upper), was dissected. There was no adhesion between the tumor and the neck of the aneurysm. The tumor was found to have sealed the bleb on the aneurysm dome, at which the aneurysm was presumed to have ruptured. A Sugita No. 1 clip was used to obliterate the aneurysm successfully (Fig. 4 lower). Residual tumor on the bleb and in the sella turcica was resected after clipping, without fear of premature rupture. The tumor was totally removed and the attachment at the tuberculum sellae was entirely coagulated (Simpson Grade 2). The pathological diagnosis was meningotheliomatous meningioma. No hemosiderin deposit was found anywhere in the specimen.

Operation. The operation was performed on the 20th day post-SAH. While the patient received a general anesthetic, a coronal skin incision was made followed by a bilateral frontal craniotomy. Via the subfrontal and interhemispheric approaches, a reddish-gray tumor was encountered between the olfactory nerves. After piecemeal resection, the bilateral optic nerves and carotid arteries were identified. Although the tumor encased the bilateral ACAs, the ACoA, and the Heubner arteries, there was no adhesion to any of these vessels. The bilateral $A_1$ was temporarily clipped and the aneurysm, which originated from the left $A_1-A_2$ junction (Fig 4. upper), was dissected. There was no adhesion between the tumor and the neck of the aneurysm. The tumor was found to have sealed the bleb on the aneurysm dome, at which the aneurysm was presumed to have ruptured. A Sugita No. 1 clip was used to obliterate the aneurysm successfully (Fig. 4 lower). Residual tumor on the bleb and in the sella turcica was resected after clipping, without fear of premature rupture. The tumor was totally removed and the attachment at the tuberculum sellae was entirely coagulated (Simpson Grade 2). The pathological diagnosis was meningotheliomatous meningioma. No hemosiderin deposit was found anywhere in the specimen.

Postoperative Course. The patient’s postoperative course was uneventful except for bilateral anosmia and normal-pressure hydrocephalus. The patient was discharged in good condition after undergoing ventriculoperitoneal shunt placement.

Discussion

The incidence of aneurysms in patients with brain tumors is reported as 0.2 to 0.7%, 6,7,15,23 which is estimated to be lower than the true incidence because four-vessel angiographic studies are not always performed in patients harboring intracranial tumors.23 The most frequently involved intracranial tumor associated with aneurysms is the meningioma.5,7,15 The first symptoms reported by these patients were related to the tumor in 78% of cases, whereas the aneurysm was the cause of the first symptoms in 22%.18 Subarachnoid hemorrhages in these patients were always secondary to aneurysm ruptures.7,13,15 As far as we know, this is the first report of an SAH caused by the rupture of an aneurysm embedded in a meningioma.

Several mechanisms of aneurysm formation associated with meningiomas have been proposed. Kandel, et al.,8 described a patient in whom a saccular aneurysm of the middle cerebral artery was enclosed in a frontotemporal meningioma. Under microscopic examination, the tumor was found to be intimately attached to the arterial adventitia. These authors ascribed development of the aneurysm to the damage to the middle cerebral artery caused by the meningioma. Although we did not examine the aneurysm wall microscopically in our case, we presumed that the tumor had not invaded the arterial wall and the aneurysm because the lesion was easily dissected from the enclosed vasculature except for the point of rupture of the aneurysm. Pia, et al.,15 attributed the mechanism to increased regional blood flow to the tumor. Scamoni et al.,18 reported that the meningioma and aneurysm were observed on the same side in more than 80% of the patients reported in the literature. In our case, however, the aneurysm did not arise on the main feeding vessel to the tumor. We therefore considered other factors such as a particular genetic background,9 stimulated vascular proliferation,10 or simple co-incidence.14,20

We chose the bilateral frontal craniotomy, which pro-
Ruptured aneurysm inside meningioma

duces a wide surgical view between the two ICAs, allowing the lesion to be approached via various routes. However, in our patient, an ordinary pterional or unilateral frontal craniotomy would have been enough to avoid damaging her olfactory nerves, because the tumor was soft and did not adhere to the encased vasculature. We usually cannot determine preoperatively whether it will be possible to dissect the tumor from the encased vasculature. Sekhar and Javed\textsuperscript{19} stated that the presence of an arachnoidal plane is inferred during surgery by the ease of dissection. Kawase, et al.\textsuperscript{9} emphasized the difficulty of dissecting meningiomas from the encased arteries without the presence of the arachnoidal plane. Al-Mefty\textsuperscript{1} classified clinoidal meningiomas into three categories according to the presence of arachnoid membranes between the tumor and cerebral vessels. In his Group 1, the tumor adheres directly to the adventitia, which prevents total removal and leads to poor outcomes. Preoperative angiographic studies demonstrate narrowing of the encased arteries in this group. In our case, however, the embedded aneurysm caused diffuse SAH rather than intertumoral hemorrhage. Preoperative angiographic studies did not reveal arterial narrowing. These factors might have indicated that a regional pial or arachnoidal layer was kept intact, which limited adhesion between the vascular structures and the tumor.

In a very complicated case such as the one reported in this article, careful stepwise procedures are essential to treat the aneurysm and the tumor simultaneously. We started with piecemeal resection of the tumor to achieve precise anatomical orientation of the encased arteries. After temporary clipping of the bilateral A1, we removed the tumor around the aneurysm neck and applied a suitable clip. Clipping the residual tumor together with the aneurysm may lead to incomplete clipping or tumor regrowth. After complete clipping of the aneurysm, we could then resect the residual tumor on the bleb of the aneurysm dome without fear of rupture.

Although early surgery is now a widely accepted treatment for aneurysms,\textsuperscript{15} we chose late surgery because we wanted to ensure that the patient, who was relatively elderly and had a complex of pathological findings, was in stable condition at the time of surgery. A two-stage treatment is also possible in such cases, but the aneurysm could only be safely clipped after the aforementioned careful stepwise tumor removal. Embolization of the aneurysm in an acute stage followed by late phase tumor removal is another alternative.

**Conclusions**

This is the first report of SAH secondary to a ruptured aneurysm completely encased in a meningioma. Preoperative angiographic studies revealed encased arteries with normal diameters. Hemorrhage from the aneurysm was seen diffusely in the subarachnoid space beside the embedded arteries. These factors indicated very little adhesion between the tumor and the encased vascular structures. We successfully removed the tumor and clipped the aneurysm on the 20th day post-SAH. Careful stepwise procedures were essential to treat the aneurysm and the tumor simultaneously.

**References**

9. Kawase T, Shiobara R, Toya S: Middle fossa transpetrosal-transventorial approaches for petroclival meningiomas: selec-

Manuscript received May 5, 1999.
Address reprint requests to: Masahiro Ogino, M.D., D.M.Sc., Department of Neurosurgery, Dokkyo University School of Medicine, 880 Kitakobayashi, Mibu, Shimotsuga, Tochigi 321-0293, Japan. email: oginom@dokkyomed.ac.jp.