Ruptured Aneurysm of the Orbitofrontal Artery Associated With Dural Arteriovenous Malformation in the Anterior Cranial Fossa
—Case Report—

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Abstract
A 27-year-old male presented with a rare association of a ruptured orbitofrontal artery aneurysm and a dural arteriovenous malformation (DAVM) fed by both ethmoidal arteries, manifestation as severe headache, nausea, and vomiting. Computed tomography revealed a hematoma within the right frontal lobe and diffuse subarachnoid hemorrhage. The aneurysm was clipped successfully and the hematoma was evacuated. After an uneventful postoperative course, the patient was referred for gamma knife radiosurgery to treat the DAVM. In this case, the DAVM was asymptomatic and pathogenetically unrelated to the aneurysm, which demanded urgent treatment.

Key words: cerebral aneurysm, arteriovenous malformation, radiosurgery

Introduction
Distal anterior cerebral artery aneurysms are uncommon, accounting for about 5% of cerebral aneurysms.5,12) Within this small subgroup, only two ruptures of an orbitofrontal artery aneurysm have been reported.2,4) Dural arteriovenous malformations (DAVM) most frequently involve the sigmoid or cavernous sinus and are less common in the anterior cranial fossa.1,6,7,10,11,15,16,18) We treated a patient with the rare association of a ruptured aneurysm of the orbitofrontal artery and a DAVM in the anterior cranial fossa.

Case Report
A 27-year-old male suddenly developed severe headache, nausea, and vomiting after a meal in December 1996 and was brought to our hospital by ambulance. No bruit was detected over the head or neck. His blood pressure was 146/78 mmHg. Computed tomography (CT) at admission suggested diffuse subarachnoid hemorrhage accompanied by hemorrhage within the right frontal lobe (Fig. 1 left). No acute subdural hematoma was seen. Left internal carotid angiography revealed an aneurysm of the left distal orbitofrontal artery with a maxi-

Fig. 1 Computed tomography scans, on admission (left) showing an intracerebral hematoma in the right frontal lobe and subarachnoid hemorrhage, and after angiography (right) showing an enhanced mass lesion in the interhemispheric fissure continuous with the hematoma.
mum diameter of 1 cm, and abnormal progression of contrast medium from the left anterior ethmoidal artery into a dilated right olfactory vein, then into the right basal vein of Rosenthal (Fig. 2). Right internal carotid angiography showed no contrast medium entered the region distal to the A\textsubscript{1} segment of the anterior cerebral artery because of the compression by the hematoma. CT following angiography suggested that the aneurysm in the interhemispheric fissure was responsible for the intracerebral hematoma in the right frontal lobe continuous with the aneurysm was evacuated. Treatment of the DAVM was deferred.

The patient had an uneventful postoperative course. He regained clear consciousness on the day following the surgery, and developed no clinically evident cerebral vasospasm. Postoperative left internal carotid angiography one month after admission suggested partial occlusion of the parent artery of the aneurysm (Fig. 3). Right internal carotid angiography suggested that the right anterior ethmoidal artery, arising from the right ophthalmic artery, was feeding the DAVM in the anterior cranial fossa. The patient was referred for radiosurgery to treat the DAVM in the right anterior cranial fossa at another hospital and was discharged ambulatory from our institution. Two months after the onset, the patient received 40 Gy directed to the DAVM by the gamma knife technique, with radiation to the marginal areas limited to 16 Gy (40%). The patient had a good overall clinical result according to regular follow-up evaluations over the next few months.

**Discussion**

Eight cases of cerebral aneurysm associated with DAVM have been reported, of which detailed information is available for five cases (Table

![Fig. 2 Preoperative left internal carotid angiograms, lateral views (left, center) and anteroposterior view (right), in the arterial phase (left, right) showing a saccular aneurysm (arrow) involving the left orbitofrontal artery and a dural arteriovenous malformation (arrowheads) fed by the left anterior ethmoidal artery, and in the late arterial phase (center) showing a dilated right olfactory vein draining the dural arteriovenous malformation.](image)

![Fig. 3 Postoperative left internal carotid angiograms, lateral view (left) and anteroposterior view (right), in the arterial phase showing occlusion of the left orbitofrontal artery, but no change in the dural arteriovenous malformation.](image)
1) 3,7,9,10,16) All patients were males, and four had intracranial hemorrhage from rupture of the aneurysm. 3,9,10,16) Aneurysm surgery was performed before treatment of the DAVM. The parent artery of the aneurysm also fed the DAVM in three cases suggesting that hemodynamic stress had contributed to development of the aneurysm. 3,9,10) The other three aneurysms7.10,16) were associated with DAVM in the anterior cranial fossa involving the anterior communicating artery, the bifurcation of the internal carotid artery, and the posterior communicating artery. The orbitofrontal artery was involved in our patient. No mechanistic association linking the DAVM to these aneurysms seems possible.

The association of aneurysm and DAVM in our case probably a chance occurrence, since the main portion of DAVM and the draining vein were located contralateral to the aneurysm. Postoperative cerebral angiography revealed partial occlusion of the distal portion of the aneurysm's parent artery, whereas the DAVM showed an appearance little changed from the preoperative angiography. However, superselective angiography of the orbitofrontal artery was not performed preoperatively, so whether the orbitofrontal artery was the artery feeding the DAVM was not established with certainty.

The aneurysm in our case was located distally to the anterior communicating artery, and can be classified as a distal anterior cerebral artery aneurysm. The most common site in this area is the bifurcation of the pericallosal and callosomarginal arteries at the genu of the corpus callosum or superiorly to this point. Distal anterior cerebral artery aneurysms were located in the frontobasal region in only eight of 92 cases. 5) Analysis of 209 cases showed that 15% occurred in the infracallosal portion, and that such aneurysms were likely to occur at the origin of the frontopolar artery. 12) Only two such aneurysms have arisen from the distal orbitofrontal artery. 4) Acute angulation of the distal orbitofrontal artery was postulated as the cause of hemodynamic stress in one case, 4) and a recurrent configuration of the anterior ethmoidal arteries as the potential cause of hemodynamic stress in the other. 2) In our case, acute angulation was present, as observed in the orbitofrontal artery. 4) Distal orbitofrontal artery aneurysms may be particularly difficult to dissect because of limited space in the interhemispheric fissure, a broad base, sclerotic plaques at the aneurysmal neck, or association with other aneurysms. 13,19)

Initial CT of the present case suggested a typical DAVM in the anterior cranial fossa, although the origin of hemorrhage was difficult to determine. Repeat CT performed following angiography verified the continuity between the hematoma and the aneurysm, indicating the aneurysm as the cause of hemorrhage. This finding was confirmed at operation, supporting the utility of postangiographic CT.

DAVM in the anterior cranial fossa associated with intracranial hemorrhage is generally treated surgically7,10,13,19,18) in the acute phase. We operated only on the aneurysm, which we considered to be the cause of hemorrhage. The need for emergent treatment of the ruptured aneurysm resulted in deferral of treatment for the DAVM. Normally, operations for DAVM in the anterior cranial fossa, such as ligation of the draining vein, 15) are not difficult. In retrospect, since the draining vein was only dilated, and not an aneurysmal sac, simultaneous treatment of these two unusual vascular abnor-
malities at one operation may have been the better choice.

Radiosurgery was chosen for the DAVM, although DAVM in the anterior cranial fossa has a relatively high risk of hemorrhage. Radiosurgery is a recognized noninvasive treatment for DAVM,\textsuperscript{1,5,11,17} and the DAVM had not caused hemorrhage or other symptoms, and no aneurysmal sac involving the draining vein (usually the cause of any hemorrhage from a DAVM) was identified angiographically.\textsuperscript{6}

References


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