

Arteriovenous Fistula Arising From the Persistent Primitive Olfactory Artery With Dual Supply From the Bilateral Anterior Ethmoidal Arteries

—Case Report—

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Abstract

A 59-year-old male presented with generalized seizure. The patient had not been aware of any traumatic head injuries or preceding infection, and had no contributory medical history. On admission, he was alert and well oriented, without neurological impairment or headache. He was afebrile and blood examination showed no abnormal findings. Computed tomography revealed an irregular intracerebral hematoma, 3 × 1.5 cm in diameter, in the left rectal gyrus. Cerebral angiography showed an arteriovenous fistula (AVF) in the anterior cranial fossa supplied only by the persistent primitive olfactory artery (PPOA) originating from the anterior cerebral artery, forming a shunt to an ascending cortical vein, and drained by the superior sagittal sinus. The patient underwent endovascular obliteration of the AVF via the transarterial route. Immediately after successful isolation, angiography showed that the bilateral anterior ethmoidal arteries supplied the AVF. The feeding branches from the left anterior ethmoidal artery were completely occluded via the ophthalmic artery, but introduction of the catheter into the right ophthalmic artery markedly decreased the stump pressure. Follow-up angiography performed at 3 and 8 weeks following embolization showed spontaneous resolution of the residual AVF without findings of recanalization or new abnormal channels. AVF arising in the anterior cranial fossa may be associated with an unusual pattern of the blood supply when including the PPOA.

Key words: dural arteriovenous fistula, anterior cranial fossa, persistent primitive olfactory artery, endovascular therapy

Introduction

Arteriovenous fistula (AVF) in the anterior cranial fossa is a rare but aggressive entity manifesting as intracerebral hemorrhage or subarachnoid hemorrhage,^{1,5,11} and usually supplied by the anterior ethmoidal artery or exceptionally by the branches of the anterior cerebral artery.^{2,3} Surgical obliteration of the abnormal shunt is the preferred therapeutic option, but recently endovascular management or radiosurgery and multidisciplinary approaches have also become potential therapeutic approaches.^{1,4-6,9,11}

The persistent primitive olfactory artery (PPOA) is a rare remnant of the fetal arteries that usually dwindles but infrequently persists as a small branch accompanying the olfactory nerve into the nasal cavity, and a larger derivative extends laterally to enter the mesial part of the anterior perforated substance as the recurrent artery of Heubner at 44 days ovulation age. The PPOA remains only as part of the recurrent artery of Heubner in normal vascular developments.⁸ The PPOA has been associated with aneurysmal subarachnoid hemorrhage, but not with AVF.^{7,12}

Here we describe a case of AVF located in the frontal fossa and initially thought to be fed only by the PPOA but was also supplied by the bilateral anterior ethmoidal arteries following initial embolization.

Case Report

A 59-year-old male suffered generalized seizure and was referred to our hospital. His past medical history was unremarkable and he was not aware of any head injuries or preceding infection. On admission, the patient was alert and well oriented, without neurological impairment or signs of increased intracranial pressure. He was afebrile and blood examination showed no abnormal findings.

Computed tomography (CT) revealed an irregular intracerebral hematoma, 3 × 1.5 cm in diameter, in the left rectal gyrus. No subarachnoid hemorrhage was identified (Fig. 1). Three-dimensional CT angiography demonstrated a vascular lesion located in the anterior cranial fossa, originating from the right anterior cerebral artery and draining into the superior sagittal sinus via an ascending cortical vein (Fig. 2). Cerebral angiography identified the vascular lesion as an AVF fed by the PPOA, arising near the junction of the first and second segments of the right anterior cerebral artery, coursing anteriorly on the lower

surface of the frontal lobe across the midline, and shunted to a frontal cortical vein at the cribriform plate, forming a venous pouch. The recurrent artery of Heubner was not identified. The ophthalmic arteries were not dilated without filling of the ethmoidal branches. External carotid angiography showed normal findings on both sides (Fig. 3).

The patient underwent endovascular exploration via the transarterial route. A guiding catheter was introduced into the feeding vessel via the anterior cerebral artery and passed close to the shunt point. Continuous injection of dilute glue (33% N-butyl cyanoacrylate [NBCA]) resulted in complete disappearance of the fistula. Angiography following obliteration revealed filling of the bilateral anterior ethmoidal arteries, predominantly on the left, supplying the AVF (Fig. 4). A microcatheter (Excior SL-10 2M; Boston Scientific Co., Natick, Mass., U.S.A.) was then introduced into the third segment of the left ophthalmic artery, followed by injection of 33% NBCA, which completely isolated the blood supply from the left side. The microcatheter was then placed in the right ophthalmic artery, but the stump pressure was markedly reduced, so fur-

ther catheterization was abandoned considering the high risk of post-procedural visual impairment.

The patient was discharged following an uneventful postoperative course. Follow-up angiography performed at 3 and 8 weeks after embolization showed spontaneous obliteration of the residual shunt without findings of recanalization or new abnormal channels (Fig. 5).

Discussion

In the present case, the AVF in the anterior fossa was first identified as arising from the PPOA. The ethmoidal arteries were not demonstrated as feeding vessels until initial embolization of the AVF. To date, only two cases of dural AVF in the cranial fossa fed by the pial vessels have been described.^{2,3)} One case manifested as subarachnoid hemorrhage, and arose from the junction of the first and second segments of the anterior cerebral artery, and connected to



Fig. 1 Axial computed tomography scan demonstrating an irregular intracerebral hematoma, 3 × 1.5 cm in diameter, in the left frontal gyrus (arrowhead), and a part of the anterior cranial base (arrow).

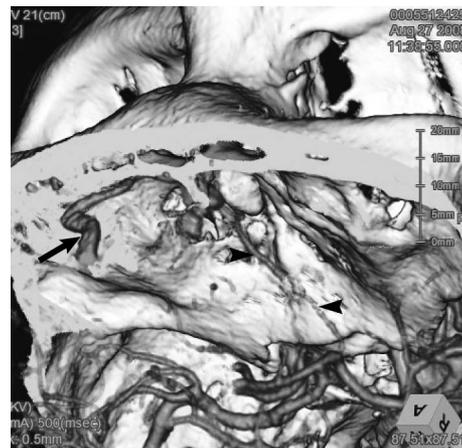


Fig. 2 Three-dimensional computed tomography angiogram, anterior superior view, showing a vascular lesion originating near the junction of the first and second segments of the right anterior cerebral artery (arrowheads), and draining into the superior sagittal sinus through an ascending cortical vein (arrow).

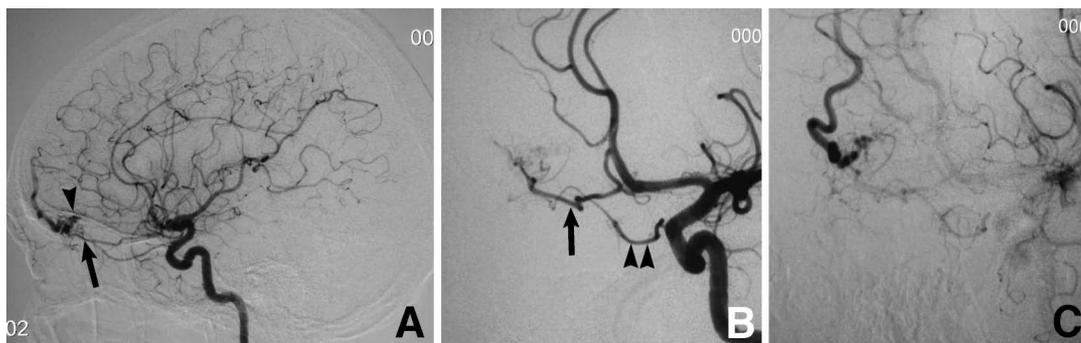


Fig. 3 Left internal carotid angiograms, lateral view (A) revealing an arteriovenous fistula (AVF) fed by the persistent primitive olfactory artery arising from the right anterior cerebral artery (arrow), forming a venous pouch (arrowhead), and oblique views in the early (B) and late (C) arterial phases demonstrating the AVF without dilation of the ophthalmic artery (arrowheads) and identifiable recurrent artery of Heubner.

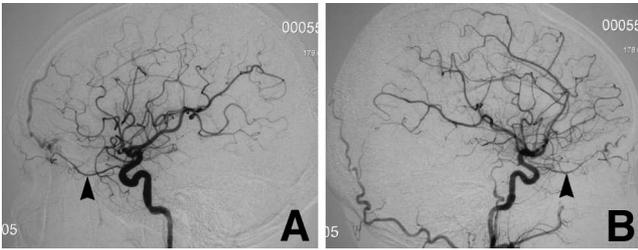


Fig. 4 Left (A) and right (B) internal carotid angiograms, lateral views, demonstrating an interrupted blood supply from the right anterior cerebral artery, and filling via the bilateral ophthalmic arteries (arrowheads), predominantly on the left.

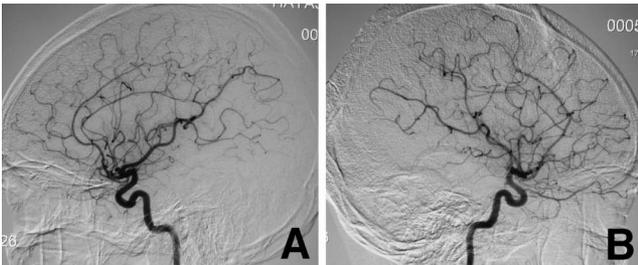


Fig. 5 Left (A) and right (B) internal carotid angiograms, lateral views, performed 3 weeks after embolization revealing spontaneous obliteration of the arteriovenous fistula.

a vein spreading over the fronto-orbital gyrus and gyrus rectus.²⁾ The other case manifested as intracerebral hemorrhage, and was supplied by both the anterior ethmoidal and frontopolar arteries, and drained by the superior sagittal sinus.³⁾ Therefore, the present case was unique in that both pial and meningeal arteries supplied the AVF, although the meningeal branches were angiographically occult until isolation of the pial supply. This dual blood supply may indicate some part of the etiology of the AVF arising in the anterior fossa.

Although the natural history of dural AVF is not fully understood, a recent retrospective study of dural AVFs with angiographically identified cortical venous reflux found that the annual risk for hemorrhage was 7.4% in patients presenting with and 1.5% in patients without intracranial hemorrhage, which were less than previously reported.¹⁰⁾ However, the cases of AVF located in the anterior fossa, which comprised 6% of the 85 cases, were not described as independent entities. Interestingly, the offending dural AVF had occluded spontaneously in 3 patients (4%) before repeat angiography performed at 9 years 8 months, 7 months, and 3 months following initial angiography. In the present case, the residual blood supply from the right ethmoidal artery ceased spontaneously within 3 weeks after embolization, followed by stable findings without recanalization or new abnormal channel confirmed at 8 weeks after embolization.

The present case was first considered to be an AVF supplied only by the PPOA originating near the junction of the first and second segments of the right anterior cerebral

artery, so was thought to be easily accessible and safe for obliteration by an endovascular procedure. In addition, the ruptured AVF was associated with less expansive intracerebral hemorrhage not causing neurological impairment. Therefore, we elected to perform endovascular therapy as first-line treatment. Any persistent residual blood supply from the right anterior ethmoidal artery was considered to be safely interrupted by consecutive open surgery. Alternatively, microsurgical interruption would have been curable for obliterating the abnormal channels formed not only within the dural, but also pial vessels. Anyway, longer follow up is needed to evaluate the final outcome of the present AVF.

The present case shows that AVF in the anterior cranial fossa may be associated with an unusual pattern of the blood supply including the PPOA.

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