

# Brain Arteriovenous Malformation Involving a Persistent Primitive Olfactory Artery



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## Abstract

The incidence of a persistent primitive olfactory artery is extremely rare. This anomaly may be involved in the development of aneurysms due to hemodynamic stress. We report a patient with a brain arteriovenous malformation mainly fed by a persistent primitive olfactory artery. A 40-year-old healthy man experienced transient numbness around the left side of his mouth. Magnetic resonance imaging incidentally disclosed flow voids inside and at the medial part of the pre-central gyrus of the left frontal lobe with dilation of the cortical veins. A left internal carotid artery angiogram revealed that a left persistent primitive olfactory artery with a proximal hair-pin turn fed an anterior component of the nidus. A right internal carotid artery angiogram showed that the right bihemispheric anterior cerebral artery fed a posterior component of the nidus. There was no aneurysm in the nidus or on the feeding vessel. The diagnosis was asymptomatic arteriovenous malformation (Spetzler-Martin grade 3). The patient developed systemic exanthema-with facial edema after diagnostic angiography. We ruled out target embolization followed by stereotactic radiosurgery and placed him under close conservative outpatient observation. We suspect that the arteriovenous malformation was due to the presence of a pre-existing persistent primitive olfactory artery. In patients with a persistent primitive olfactory artery, structural fragility and hemodynamic stress may elicit aneurysms at the apex of the hairpin curve of the persistent primitive olfactory artery.

**Key words:** Bihemispheric anterior cerebral artery, Brain arteriovenous malformation, Persistent primitive olfactory artery.

## Introduction

The primitive internal carotid artery (ICA) diverges in a rostral and caudal direction. The rostral portion supplies telencephalon including rhinencephalon, and the caudal one supplies diencephalon and mesencephalon. The

persistent primitive olfactory artery (pPOA) originates at the rostral portion of the primitive ICA, extends towards the nasal fossa at five weeks of gestation and transiently supplies the nasal cavity along the olfactory tract. The medial and lateral olfactory arteries (OAs) develop from the POA; in the final process of development, the medial OA constitutes future anterior cerebral artery (ACA) and the lateral OA becomes the recurrent artery of Heubner, the anterior choroidal artery, and the middle cerebral artery.<sup>1</sup> At that time, the characteristic hairpin turn of the medial OA regresses and takes on the shape of the normal ACA. However, when this regression does not occur, a pPOA with a hairpin turn remains.

The incidence of a pPOA is only 0.14%.<sup>1,2</sup> Komiyama et al.<sup>3</sup> found that 11 of 29 patients with a pPOA with a hairpin turn developed aneurysms (37.9%); in 9 of these patients the aneurysm arose at the apex of the hairpin curve. This suggests that hemodynamic stress at that site is implicated in the development of these aneurysms.<sup>4,5</sup> To the best of our knowledge, ours is the first documentation of a brain arteriovenous malformation (b-AVM) associated with a pPOA. We report a rare patient with a b-AVM that was mainly fed by a pPOA branching off the ACA.

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Case Report

A 40-year-old man with no relevant family history suddenly experienced deafness in the past. A few days before visiting us he noticed transient numbness around the left side of his mouth. Magnetic resonance imaging showed flow voids inside and in the medial part of the left frontal lobe and dilation of the cortical veins (Fig. 1A). Magnetic resonance angiography revealed a b-AVM that was fed by the bilateral ACA and drained mainly into the superior sagittal sinus (Fig. 1B). No aneurysm was detected in the nidus or on the feeding arteries. A right ICA angiogram showed that the right ACA was bihemispheric and supplied the posterior component of the nidus in the

left frontal lobe (Fig. 1C). A left ICA angiogram revealed that the left ACA fed the anterior component of the nidus. The proximal portion of the ACA looked anomalous; its course was anteromedial and it formed a posterosuperior hair-pin turn. We considered it to be a pPOA (Fig. 1D) and made a diagnosis of asymptomatic AVM (Spetzler-Martin grade 3) fed mainly by the left ACA via the pPOA and partially by the right ACA.

Target embolization followed by stereotactic radiosurgery was ruled out by his allergic reaction to the iodine contrast medium prohibited treatment and he is being observed carefully. In the course of 23-month follow-up he developed no clinical symptoms and additional imaging studies revealed no changes.

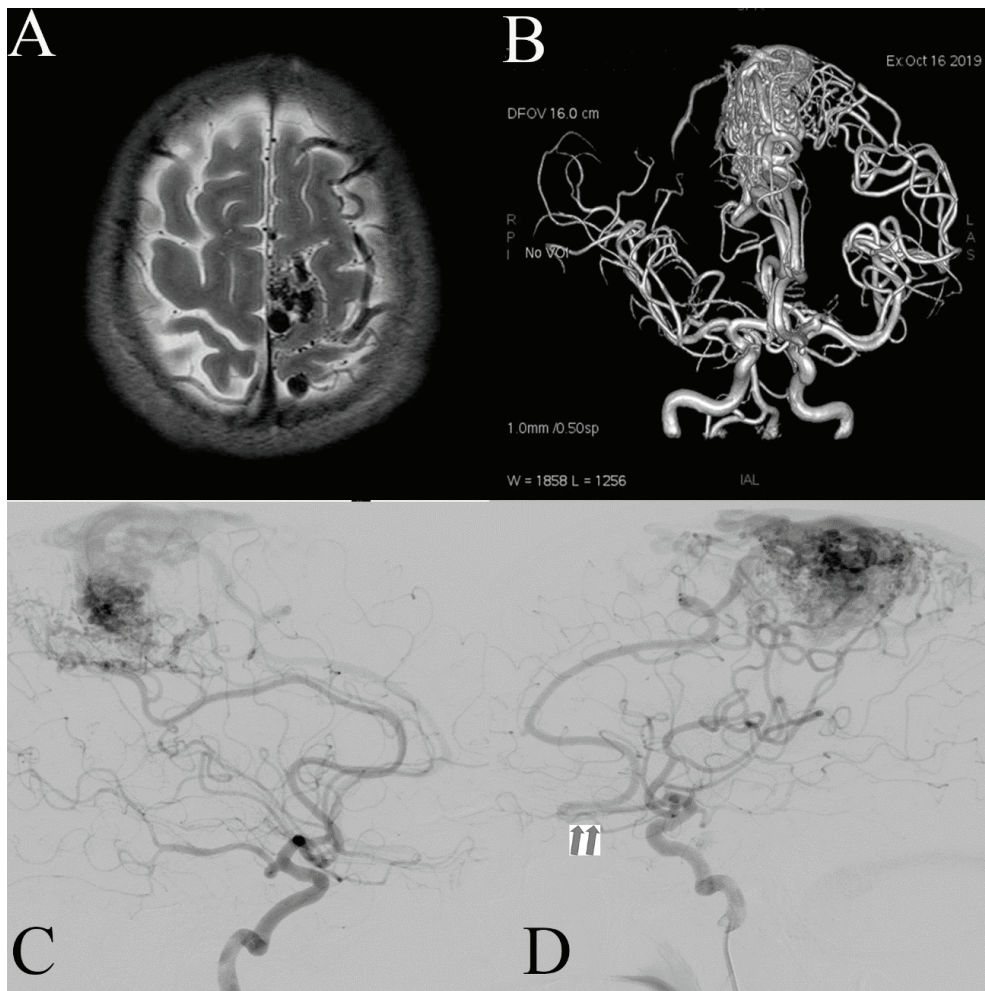


Figure 1: (A) Brain MRI (axial T2 weighted image) showing flow voids in the superior frontal- and the pre-central gyrus of the left frontal lobe with dilation of the cortical veins. (B) Magnetic resonance angiogram revealing a b-AVM fed by the bilateral anterior cerebral artery (ACA) and mainly draining into the superior sagittal sinus. (C) Right internal carotid artery angiogram demonstrating right ACA was bihemispheric and fed the posterior component of the nidus in the left frontal lobe. (D) Left internal carotid angiogram showing that the left ACA fed the anterior component of the nidus. The proximal portion of the vessel (arrows) was anomalous, and ran anteromedially with a posterosuperior hair-pin turn. It was diagnosed as a persistent primitive olfactory artery.

## Discussion

To the best of our knowledge, ours is the first report of a b-AVM fed by a pPOA. Our patient's b-AVM was supplied by the bilateral ACA, one portion was a pPOA and the other a bihemispheric ACA.

While b-AVMs fed by the ACA have been reported, b-AVMs supplied via a pPOA are very rare. We think that there is no direct association between a pPOA and b-AVMs. However, in our patient we suspect that the pPOA played a role in the development of his b-AVM.

The bihemispheric ACA is a unilateral pericallosal branch that supplies the bilateral cerebral hemispheres; its incidence is 64%.<sup>6</sup> Most of the vessel supplies only a small part of the ACA territory.<sup>6</sup> Occlusion of the distal branch of ACA during gestation may lead to the development of a bilateral ACA to augment the blood supply.<sup>7</sup> In 11 of 12 cadavers with a bihemispheric ACA, the vessel tended to supply the superior- and in 8 the inferior internal parietal artery.<sup>8</sup> In our patient, the bihemispheric ACA supplied the superior- and the inferior internal parietal arteries; they contributed as feeders to the nidus. The presence of the AVM with high flow shunts may have triggered the development of the bihemispheric ACA due to the increased demand of blood.

Due to congenital fragility and hemodynamic stress, a pPOA may elicit the development of an aneurysm at the apex of the hairpin turn.<sup>4,5</sup> In our patient, hemodynamic stress on the pPOA was increased because the vessel contributed to the b-AVM as a feeder. Flow-related aneurysms are most likely to develop around the apex of the hairpin curve of the pPOA.

When embolization is considered, the ONYX™ embolization agent is not optimal because it requires a high traction force upon retrieval of the microcatheter, which can be trapped when the plug-and-push technique is applied.<sup>9</sup> This may lead to hemorrhagic complications due to the deformity and dislocation of the hairpin-shaped vessel. We prefer N-Butyl-2 cyanoacrylate because it allows insertion of a softer catheter such as the Magic™ catheter which is more easily navigated through tortuous vessels without a guidewire and requires less traction for retrieval unless the catheter tip is captured by the glue N-Butyl-2 cyanoacrylate.

We did not perform embolization because our patient was allergic to iodine contrast media. Other treatment options are stereotaxic radiosurgery or direct surgery without embolization. We did not choose stereotaxic radiosurgery because the nidus was large and the shunt was a high flow shunt. On the other hand, surgical resection without embolization seemed to us to be too invasive for an unruptured asymptomatic AVM. Therefore, after consulting the ARUBA study<sup>10</sup> and after thorough discussion with the patient and our treatment team, we

decided to follow this patient conservatively. As he is at high risk for developing a flow-related aneurysm, we are following him closely.

## Conclusion

We encountered a patient with a very rare b-AVM fed by a pPOA. As endovascular or surgical intervention was counter-indicated, we are treating him conservatively and are following him closely.

**Conflict of Interest:** None

**Source(s) of support:** None

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