

## Cerebral Aneurysm Associated with Persistent Primitive Olfactory Artery Aneurysm

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### Summary

Persistent primitive olfactory artery is a rare vascular anomaly but has a clinical importance because of its high association with cerebral aneurysm. We describe a patient with bilateral persistent primitive olfactory arteries associated with an unruptured saccular aneurysm on the left persistent primitive olfactory artery. Seven reported cases with this anomalous artery including ours are reviewed and classified into two variants. This anomalous artery arises from the terminal portion of the internal carotid artery and courses anteromedially along the ipsilateral olfactory tract and makes a hair-pin curve posterior to the olfactory bulb, becoming the distal anterior cerebral artery (variant 1) or the ethmoidal artery (variant 2). Out of 7 reported cases, 4 cases are associated with saccular aneurysms. The aneurysm in variant 1 is located on the hair-pin curve at which an apparent arterial branch is sometimes absent. Two patients suffer from anosmia. Persistent primitive olfactory artery should be kept in mind because of its high association with intracranial saccular aneurysms and unique clinical presentation.

*Keywords:* Anterior cerebral artery; cerebral aneurysm; primitive olfactory artery.

### Introduction

Most of distal anterior cerebral artery (ACA) aneurysms are located on the bifurcation of the callosomarginal artery, and some of them are associated with vascular anomalies such as azygous ACA [4], interoptic course of ACA [7], supreme anterior communicating artery [11], carotid ACA anastomosis [6]. We report a rare case of an unruptured saccular aneurysm on the persistent primitive olfactory artery. Literature review discloses only six patients with this anomalous artery. The embryogenesis of this anomalous artery is discussed and its clinical importance is stressed.

### Case Report

A 54-year-old woman with a 3-year history of left-sided anosmia presented to the neurosurgical clinic at Kyoto University Hospital. Cerebral angiography revealed bilateral anomalous arteries arising from the terminal portion of the internal carotid artery and running anteromedially towards the cribriform plate and then abruptly turning posteriorly (hair-pin curve) to become the distal anterior cerebral arteries (Fig. 1). A saccular broad-necked aneurysm was detected at the hair-pin curve of the left anomalous artery with no apparent arterial branch at the aneurysmal site. The anterior communicating artery was not identified by carotid compression study and bilateral posterior communicating arteries were well developed (Fig. 2). The recurrent artery of Heubner was not identified on either side. Three dimensional computed tomographic angiography (3D CT angiography) also revealed the aneurysm above and bilateral anomalous arteries coursing along the olfactory tract towards the cribriform plate with no anterior communicating artery or recurrent arteries of Heubner (Fig. 3).

A bifrontal craniotomy was performed, and the aneurysm was exposed through an interhemispheric approach. The anomalous arteries on both sides were confirmed to run along the ipsilateral olfactory tract and make an abrupt turn posterior to the olfactory bulb. The left anomalous artery had the aneurysm at the hair-pin curve near the left olfactory bulb where no arterial branch was observed. The aneurysm had a broad neck and protruded anteromedially across the midline through the posterior margin of the falx cerebri. The aneurysmal neck was clipped with a curved titanium clip and coated with a piece of muscle and Biobond (Fig. 4). The postoperative course was uneventful and follow-up angiograms revealed the obliteration of the aneurysm. She was discharged from our hospital two weeks after the operation with no further neurological deficit.

### Discussion

The anomalous artery described above is characteristic in that it arises from the terminal portion of the internal carotid artery, runs along the olfactory tract towards the olfactory bulb, makes an abrupt

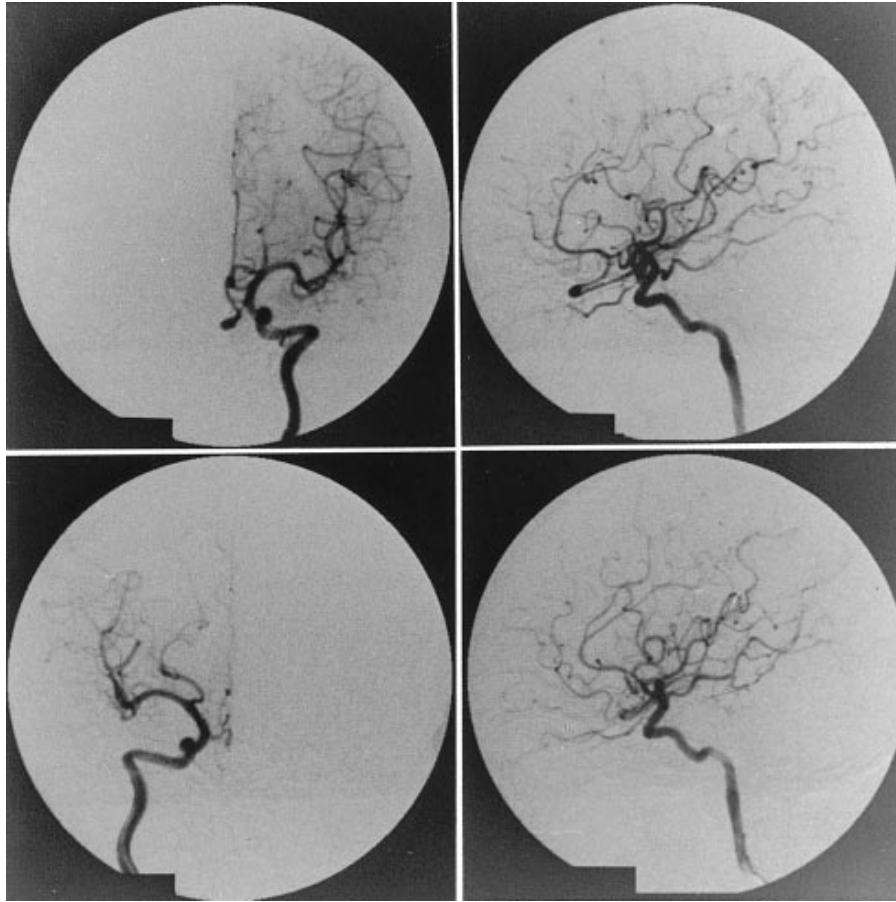


Fig. 1. Pre-operative left (upper) and right (lower) internal carotid angiograms showing anomalous arteries arising from the internal carotid arteries and run anteromedially along the frontal base and making an abrupt turn (hair-pin bend) posteriorly to become the distal anterior cerebral arteries, and a saccular aneurysm on the hair-pin bend of the left anomalous artery

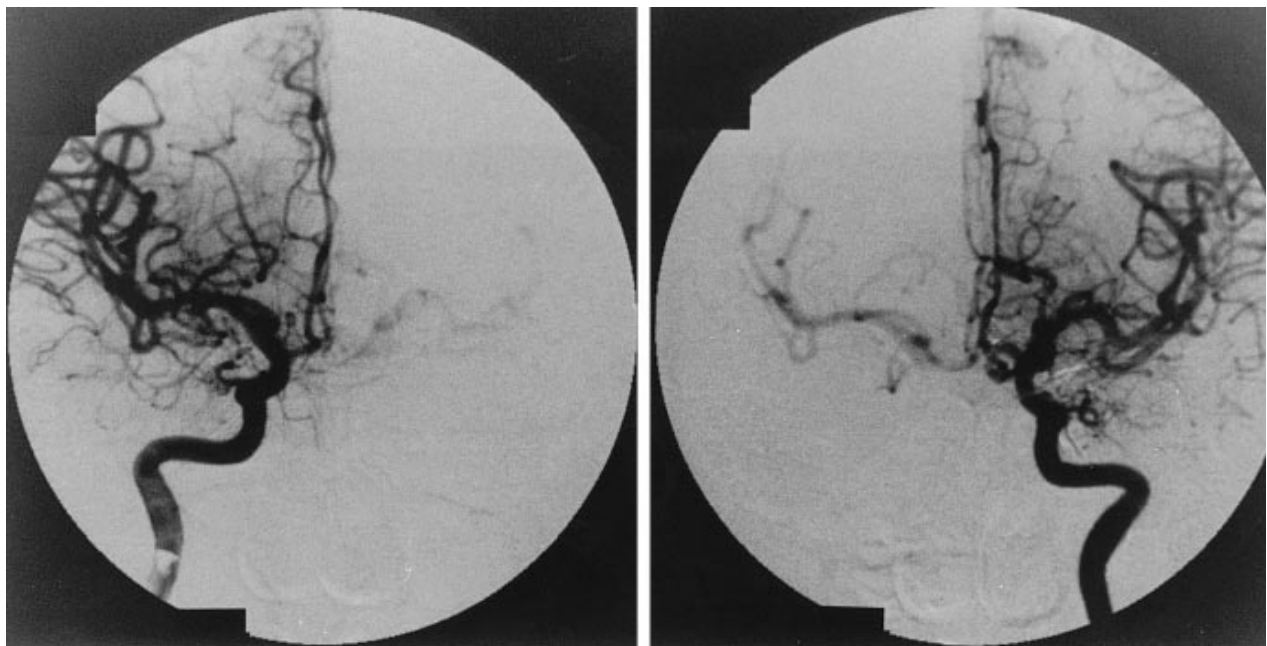


Fig. 2. Anteroposterior views of bilateral internal carotid angiograms during the contralateral carotid compression showing the absence of anterior communicating artery and recurrent arteries of Heubner. Note that contralateral middle cerebral arteries are filled via posterior communicating arteries bilaterally

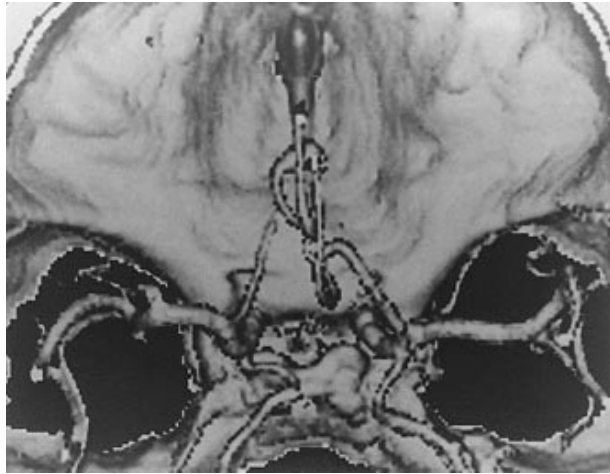


Fig. 3. Three dimensional computed tomographic angiogram showing anomalous arteries bilaterally along the frontal base and a saccular aneurysm on the left anomalous artery posteriorly to the crista galli. Note the absence of the anterior communicating artery

posterior turn behind the olfactory bulb, finally becomes the distal anterior cerebral artery, and is associated with the absence of anterior communicating artery and recurrent artery of Heubner. This anomalous artery is considered as the persistent primitive olfactory artery.

*Embryogenesis*

In the human embryo, the primitive olfactory artery appears as the cranial division of the internal carotid artery at 29 days ovulation age, and gives off the original branch to the nasal fossa and another branch more mesially toward the emerging olfactory nerve root [7]. The later branch forms the anterior cerebral artery at 35 days ovulation age. Both anterior cerebral arteries course up between the cerebral hemispheres and join with its fellow of the opposite side by plexiform anastomoses at 40 days ovulation age, which form the future anterior communicating artery. The original primitive olfactory artery dwindles but persists as a small mesial 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. Finally, the primitive olfactory artery remains only as part of the recurrent artery of Heubner in normal vascular developments. We assume that the absence of a normal proximal anterior cerebral artery, anterior communicating artery and recurrent artery of Heubner observed in the present case reflect inappropriate developments of the anterior cerebral artery and the persistence of the primitive olfactory artery.

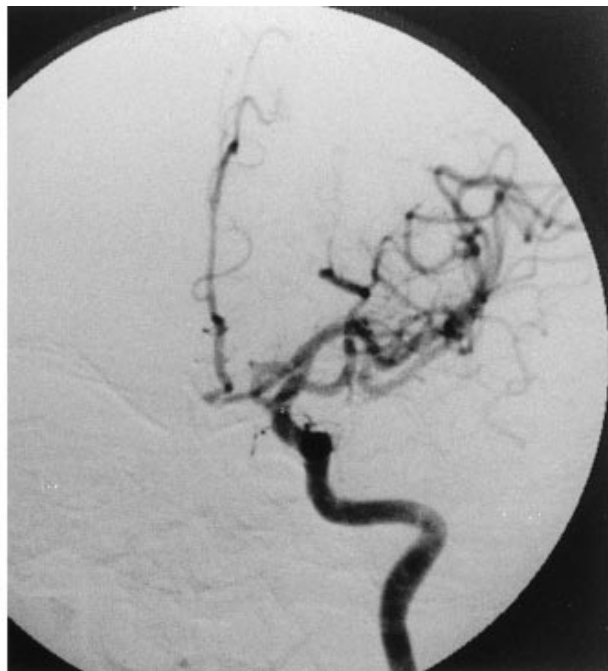


Fig. 4. Left internal carotid angiograms (left anterior oblique view) before the operation (left) and after neck clipping (right) showing the obliteration of a saccular aneurysm on the left anomalous artery

Table 1. Reported Cases of Persistent Primitive Olfactory Artery

Author's name (year)	Age/Sex	Clinical presentation	Side	Confirmation	Associated anomaly
<i>Variant 1</i>					
Takeshita (1988)	62/F	rt frontotemporal oligodendroglioma	bilateral	angiography	A com (-)
	40/F	intracerebral multiple tuberculoma	right	angiography	A com (-)
Hanakita (1988)	72/M	headache	left	angiography operation	unruptured aneurysm
Tsuji (1995)	59/F	bilateral anosmia SAH	left	angiography operation	ruptured aneurysm recurrent artery (-) long A com
Present case	54/F	left anosmia	bilateral	angiography operation	unruptured aneurysm recurrent artery (-) A com (-)
<i>Variant 2</i>					
Moffat (1967)	71/M		left	autopsy	A com (+)
Enomoto (1986)	44/M	SAH left frontal intracerebral haematoma	left	angiography operation	ruptured aneurysm

See text for Variant 1 and 2.

### Review of the Literature

A persistent primitive olfactory artery is extremely rare and literature reviews disclose only six similar cases [1, 3, 5, 9, 10]. The seven cases including ours are summarized in Table 1. The anomalous artery was confirmed only by cerebral angiography in two cases, by cerebral angiography and operation in four cases, by autopsy in one case, and located on the left side in four cases, on the right side in one case, on both sides in two cases.

These seven cases with persistent primitive olfactory arteries can be divided into two variants by their anatomical features (Fig. 5). In five cases, the anomalous artery arose from the internal carotid artery, ran along the olfactory tract and made a hair-pin bend to supply the distal anterior cerebral artery territory (variant 1). In two other cases, the artery arose from left anterior cerebral artery and passed through the cribriform plate to supply the nasal cavity as the ethmoidal artery (variant 2). Associated vascular anomalies were variable in each case, but an anterior communicating artery was absent in three cases. In one case, a long anterior communicating artery was observed. The absence of the recurrent artery of Heubner was demonstrated in two cases.

This vascular anomaly seems asymptomatic, but in two cases including ours, anosmia was observed. Because the olfactory nerve was apparently intact from

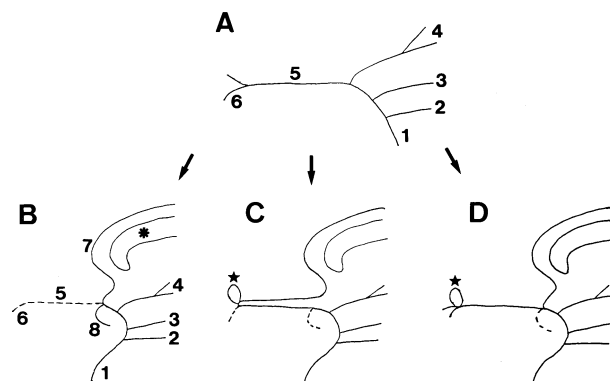


Fig. 5. Schematic drawings of the normal development of the anterior cerebral artery in human embryos (A-B) and two variants of the persistent primitive olfactory artery (C, D). A: Human embryo at 35 days of ovulation age, B: Human adult, C: Variant 1, D: Variant 2. 1 internal carotid artery, 2 posterior communicating artery, 3 anterior choroidal artery, 4 middle cerebral artery, 5 primitive olfactory artery, 6 ethmoidal artery, 7 anterior cerebral artery, 8 recurrent artery of Heubner, \*Corpus callosum. Star: cerebral aneurysm located on persistent primitive olfactory artery. Dashed lines mean the absence of the artery

the operative findings in both cases, arterial blood supply to the olfactory nerve might be involved in its pathogenesis. This vascular anomaly should be included in differential diagnosis in patients with persistent anosmia.

### *Association with Cerebral Aneurysm*

This anomalous artery is often associated with cerebral aneurysms. Cerebral aneurysms were found in four cases and two out of these four cases suffered from subarachnoid haemorrhage. Three aneurysms were located at the hair-pin bend, and two aneurysms were unrelated to the arterial bifurcation. The aneurysms usually have broad necks. The pathogenesis of cerebral aneurysms is not fully clarified, and both genetic and haemodynamic factors are considered. The hair-pin bend location of the aneurysm may imply the importance of haemodynamic stress in the occurrence of cerebral aneurysms [2]. Follow-up study should be necessary in our case to confirm whether another aneurysm will occur on the opposite persistent primitive olfactory artery. The persistent primitive olfactory artery may be overlooked because of its rarity, but a high incidence of cerebral aneurysms should be kept in mind in clinical situations.

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### Comments

The paper of our Japanese colleagues is indeed very important, as the authors call our attention to the fact that some aneurysms of

the anterior communicating artery may be related to the primitive olfactory artery.

In my series of aneurysms I did not recognise any similar occurrence. After studying the paper of the authors I am sure that I did not pay adequate attention to this anatomical variation. However, I observed during surgical exploration of the circle of Willis, regular dural branches originating from the supero-medial wall of the ICA and occasionally distal A<sub>1</sub> segment and extending to the dura of the tuberculum sellae. (See pp. 70–71, Fig. 47a–F, Vol. I Micro-neurosurgery, Thieme, Stuttgart, 1984).

Dr. Nozaki and his collaborators have presented a very concise and illustrative case report along with literature review on cerebral aneurysm associated with persistent primitive olfactory artery. This is a rare vascular anomaly but every neurosurgeon must be aware of it.

Upon reviewing this paper, there are 2 points of concern:

1) In the *Discussion* the authors have stated that the recurrent artery enters the mesial part of the anterior perforated substance.

According to Dr. Rhoton's publication on microsurgical anatomy of the anterior perforating arteries [1], the branches of the recurrent artery enter predominantly the anterior half of the anterior perforated substance along its full mediolateral extent, from the inter-hemispheric fissure to the limen insulae.

Observations from our daily surgical practice and laboratory dissections have shown that in most cases the recurrent artery enters even at the lateralmost aspect of the anterior perforated substance, therefore contrary from what was stated by the authors.

2) Our interpretation on Fig. 2 is quite different from what authors have reached:

a) The recurrent artery of Heubner is usually not visualized in internal carotid angiograms whether the compression of the contralateral carotid is applied or not, therefore, the conclusion based solely on the internal carotid angiogram that the recurrent artery is absent is at least questionable.

b) The same criticism is also extended to the statement that the anterior communicating artery is absent on the same angiogram.

c) It is extremely difficult to imagine the filling of the contralateral middle cerebral artery via bilaterally well developed posterior communicating arteries for the following reason:

If the posterior communicating arteries are well developed, it should be that of foetal type, and the vertebrobasilar system should be filled via the carotid system, so we should be able to visualize both posterior cerebral arteries by carotid injection. However we can only visualize the posterior cerebral artery on one side; why does blood go to the opposite posterior communicating artery to the opposite carotid system instead of going to the opposite posterior cerebral artery?

It is more logical to presume that there is an anterior communicating system to explain the findings of Fig. 2. This is a very concise and illustrative case report of a rare vascular anomaly, however, for the sake of publication, it would be judicious if the above mentioned questions could be answered.

### Reference

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*E. de Oliveira*

### *Authors' Reply*

1) According to Dr. Rhoton's publication . . . ,

The reviewer is right, but in our manuscript we stated the embryological development of recurrent artery according to

Padgett's publication on embryology of cranial arteries. The discussion about the location of the entrance of recurrent artery is beyond the case report, we may change the sentence on page 5, line 17, "enter the mesial part of the anterior perforated substance" into "enter the anterior perforated substance".

2) About the interpretation on Fig. 2

a), b), c) About the absence of anterior communicating artery and recurrent artery of Heubner

The interpretation on Fig. 2 is difficult and we can understand the reviewer's interpretation. We have also stated in the manuscript page 4, line 1, "The recurrent artery of Heubner were not identified bilaterally (on angiogram)". This is why we performed 3D CT angiography to add further information on the absence of anterior communicating artery and recurrent artery of Heubner.

3D CT angiography also showed no anterior communicating artery or recurrent artery of Heubner. If the anterior communicating artery is present, internal carotid angiogram with the compression of the contralateral carotid may reveal bilateral anterior cerebral artery before filling the contralateral middle cerebral artery. However, Fig. 2 shows only ipsilateral anterior cerebral artery with contralateral middle cerebral artery. Indeed, Fig. 3 shows bilateral posterior cerebral arteries. (Please note the faint vessel above the middle cerebral arteries.)

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