Case Report

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Persistent Primitive Olfactory Artery Type 4 with Fusiform Aneurysm: A Case Report

방추형동맥류를 동반한 제4형 잔류 원시 후각동맥의 영상 소견: 증례 보고

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The persistent primitive olfactory artery (PPOA) is a rare variant of the anterior cerebral artery, first reported in 1979. It reportedly has a high correlation with the development of aneurysms, owing to the hemodynamic stress induced by the structural characteristics of the hairpin turn. Herein, we present a rare case of PPOA type 4 with a fusiform aneurysm at the hairpin turn segment in a 46-year-old female with occasional headaches. Time-of-flight MR angiography and transfemoral cerebral angiography revealed an unusual branch arising from the left A1 segment, running anteromedially along the ipsilateral olfactory tract, and turning the hairpin posterior to the olfactory bulb. This branch continued into the left accessory middle cerebral artery, and a fusiform aneurysm was observed at the hairpin segment. No further treatment was performed, and follow-up imaging was recommended. Nevertheless, it is essential to recognize and diagnose these rare variations.

Index terms Arteries; Intracranial Aneurysm; Cerebral Arteries; Magnetic Resonance Angiography; Cerebral Angiography

INTRODUCTION

The persistent primitive olfactory artery (PPOA) is a rare variant of the anterior cerebral artery (ACA) that was first described by Yamaura et al. in 1979 (1). Its incidence is 0.14% in Japan (2011) and 0.29% in Korea (2012) (2). It is mostly unilateral and rarely bilateral; moreover,

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THE KOREAN SOCIETY of
RADIOLOGY

it has no laterality or sex predilection (3). According to Kwon et al. (2), the left side has a higher frequency in Korea. Five types of PPOA have been reported, and type 6 has been recently suggested (4, 5). We report a case of PPOA type 4 with a fusiform aneurysm in the hairpin turn segment.

CASE REPORT

A 46-year-old female presented with occasional headaches for 1 month. She had no specific medical history other than hypertension, diabetes mellitus. Physical examination revealed no neurological deficits, including anosmia or visual disturbances.

The patient underwent brain MRI, which revealed no abnormal findings in the brain parenchyma. Time-of-flight MR angiography revealed a saccular ophthalmic aneurysm in the left internal carotid artery (ICA). The A1 segment, an unusual branch arising from the left ACA, was also observed (Fig. 1A).

Transfemoral cerebral angiography revealed that this branch moved anteromedially along the ipsilateral olfactory tract, with a hairpin turn posterior to the olfactory bulb. Without anastomosis to the distal ACA, it continued to the left accessory middle cerebral artery (MCA). Moreover, a fusiform aneurysmal dilatation involving a hairpin curve was observed (Fig. 1B).

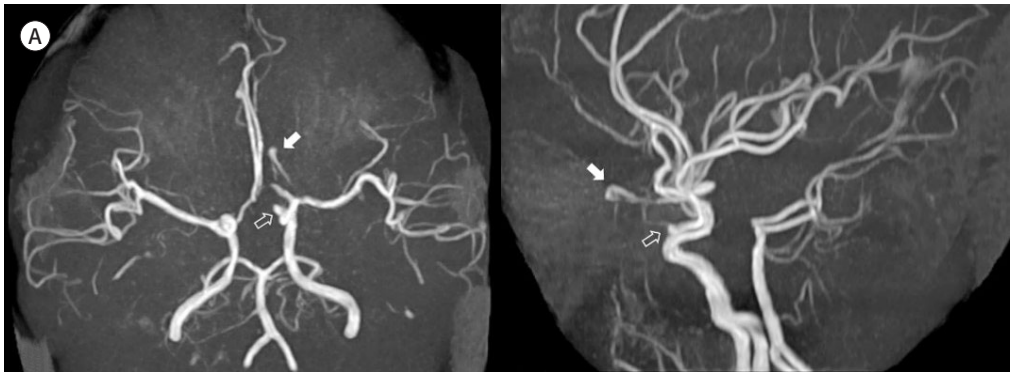
Therefore, left PPOA type 4 with fusiform dilatation was diagnosed, and no surgical or endovascular treatment was performed. Follow-up imaging is advised to monitor aneurysms.

This retrospective study was approved by the Institutional Review Board of the Inje University Busan Paik Hospital, which waived the requirement for informed consent (IRB No. 2022-10-010).

DISCUSSION

Embryologically, at 4 weeks of gestation, the rostral division from the primitive ICA be-

Fig. 1. Persistent primitive olfactory artery type 4 with aneurysm.
A. Time-of-flight MR angiography. Maximal intensity projection superoinferior view (left image) and lateral view (right image). There is a paraolfactory hairpin curve of the left PPOA (arrows). Paraophthalmic aneurysm is also noted in the left internal carotid artery (empty arrows).
PPOA = persistent primitive olfactory artery



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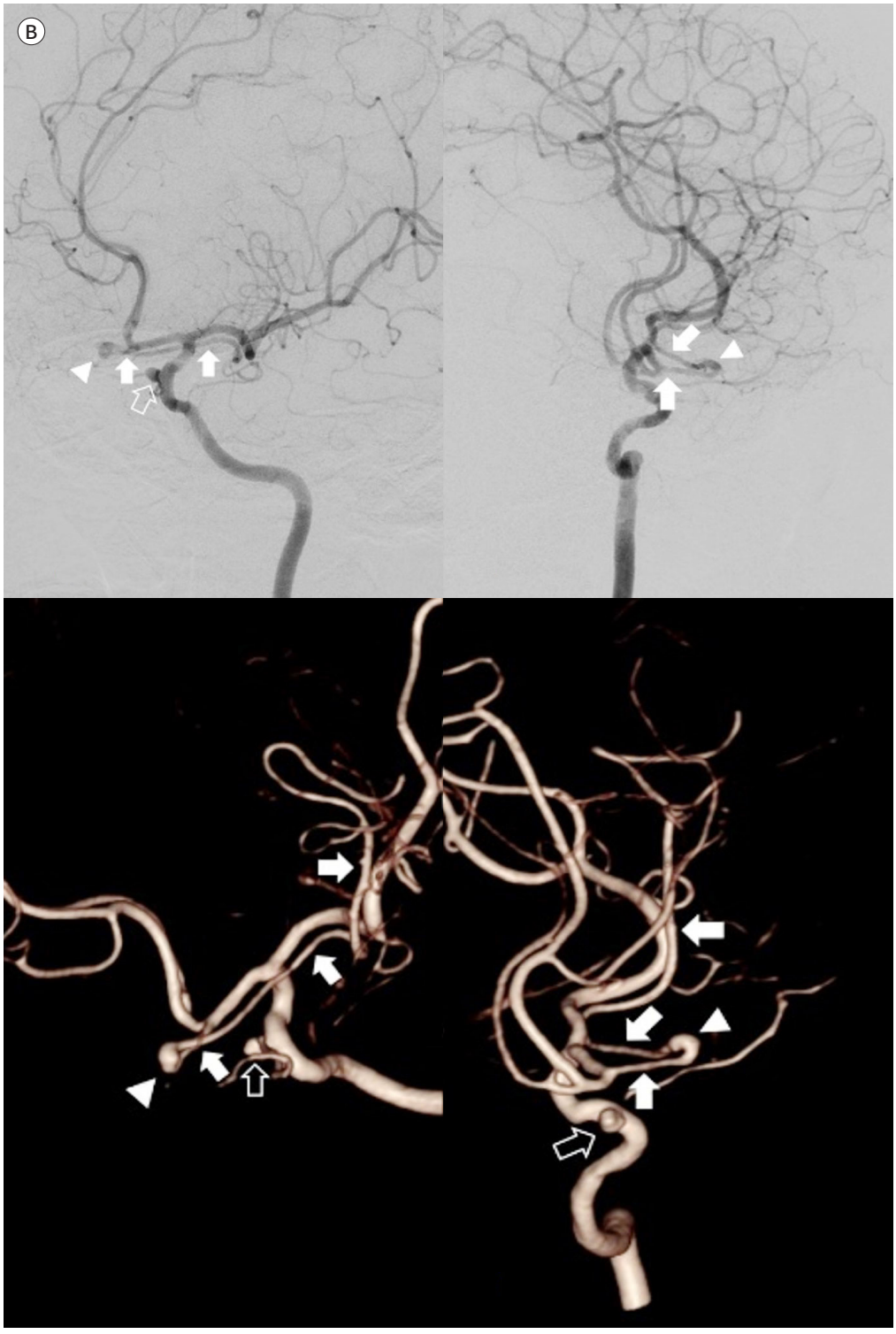
THE KOREAN SOCIETY of
RADIOLOGY

Fig. 1. Persistent primitive olfactory artery type 4 with aneurysm.
B. Digital subtraction angiography (upper images) and matched three-dimensional CT angiography (lower images) of the left ICA. Lateral view (left images) and oblique views (right images). The left PPOA arises from left A1, makes a hairpin turn, and finally continues to the left accessory middle cerebral artery (arrows). There is fusiform dilatation at the hairpin segment (arrowheads). A paraophthalmic aneurysm is also noted in the left ICA (empty arrows).
ICA = internal carotid artery, PPOA = persistent primitive olfactory artery



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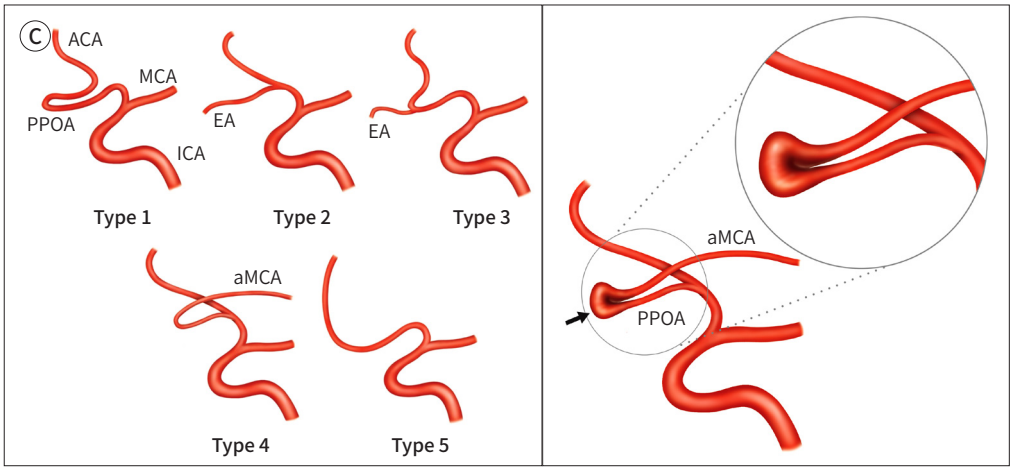
THE KOREAN SOCIETY of
RADIOLOGY

Fig. 1. Persistent primitive olfactory artery type 4 with aneurysm.
C. Illustration of five types of PPOA in the left lateral view. It is divided into five types depending on the presence or absence of the following blood vessels and hairpin turns (left image). Illustration of PPOA type 4 with hairpin-turn aneurysm (arrow) (right image).
ACA = anterior cerebral artery, aMCA = accessory middle cerebral artery, EA = ethmoidal artery, ICA = internal carotid artery, MCA = middle cerebral artery, PPOA = persistent primitive olfactory artery



comes the primitive olfactory artery (POA) with three branches. During normal development, the medial branch becomes the ACA proper, and the lateral branch becomes the recurrent artery of the Heubner, anterior choroidal artery, and MCA, but the terminal portion of the POA in the nasal fossa degenerates at around 7 weeks of gestation. However, if the regression of the terminal portion of the POA does not occur, the remnant artery PPOA persists (6).

Until recently, the PPOA was classified into five types depending on the artery that continued and whether the hairpin turn was present (Fig. 1C). Most cases reported that PPOAs are type 1, which is considered to have the most typical appearance, with other types rarely reported (5).

Type 1 arises from the proximal ACA and continues to the distal ACA, making a hairpin turn along the olfactory bulb (7).

Type 2 does not have a hairpin turn and continues into the ethmoidal artery of the nasal cavity (7).

Type 3 makes a hairpin turn and continues to the distal ACA, like type 1, with branching continuing to the ethmoidal artery, similar to type 2 (4).

After an abrupt hairpin turn, type 4 continues into the accessory MCA (4).

Type 5 does not have hairpin turns and continues to the A3 segment of the ACA (4).

Clinically, PPOA is strongly associated with the development of aneurysms. Several cases of subarachnoid hemorrhage (SAH) with ruptured aneurysms in the hairpin turn segments have been reported (3, 7, 8). Although the exact pathogenesis has not yet been elucidated, hemodynamic characteristics induced by the curvature of the abrupt hairpin turns are thought to play a role in this phenomenon (Fig. 1C). Two of the 29 patients with PPOA and one of the 14 patients in the studies by Kwon et al. (2) and Uchino et al. (3) had hairpin-turn segment aneurysms.

Another clinical presentation of PPOA is anosmia. There have been several case reports of

anosmia, some of which were unilateral or bilateral, and some patients recovered postoperatively (2, 7, 8). According to Kwon et al. (2), it was reported that two of the 28 (7.1%) patients with PPOA had anosmia. This was thought to be related to olfactory nerve perfusion.

PPOA is mostly found incidentally but can also be found as a neurologic defect or SAH, as described above. Therefore, even if a clinically asymptomatic PPOA is noted, regular examinations, including computed tomography angiography, are required to monitor the aneurysm. Moreover, these arterial variants are important in intracranial surgeries of the skull base to prevent vascular complications.

In conclusion, we found a rare variant of PPOA linked to the accessory MCA rather than to the distal ACA. Type 4 PPOA has been classified relatively recently and has been reported in only a few cases compared to the classification of other types of PPOA (9). It is important to note the different types of PPOA and identify the accompanying symptoms that may occur.

Author Contributions

Conceptualization, B.J.W.; data curation, P.H., B.J.W.; formal analysis, P.H., H.Y.J.; investigation, P.H., Y.S.; methodology, H.J.; project administration, P.H., B.J.W.; resources, P.H., J.H.W.; supervision, B.J.W., J.H.W.; validation, B.J.W., J.H.W.; visualization, P.H.; writing—original draft, P.H., B.J.W.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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방추형동맥류를 동반한 제4형 잔류 원시 후각동맥의 영상 소견: 증례 보고

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잔류 원시 후각동맥은 1979년에 처음 보고된 매우 드문 전대뇌동맥의 변이로, 급격하게 꺾이는 머리핀 회전의 구조적 특성에 의하여 혈역학적 스트레스가 유발되고, 이로 인하여 동맥류의 발생과 높은 연관성을 가지는 것으로 보고되고 있다. 우리는 간헐적 두통을 주소로한 46세 여성에서 우연히 발견된 머리핀 회전에서 동맥류를 동반한 제4형 잔류 원시 후각 동맥의 증례에 대하여 보고하고자 한다. 뇌 MRA와 유체속도강조 자기공명혈관조영술(time-of-flight MR angiography)에서 왼쪽 전대뇌동맥의 A1 분절에서 시작되어 머리핀 회전을 형성한 후 부 중대뇌동맥으로 이어지는 비정상적인 주행을 보이는 동맥이 확인되었다. 또한 머리핀 회전 분절에서 방추형 동맥류도 확인이 되었다. 이러한 변이들은 극히 드물긴 하지만, 동맥류가 동반될 수 있음을 인지하고 진단하는 것이 중요하다.

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