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Persistent Trigeminal Artery Variant Terminating in the Ipsilateral Superior Cerebellar Artery 상소되동맥영역을 공급하는 지속성 원시 삼차신경동맥 변이

Tae Young Park, MD ^(D), Byung Hoon Lee, MD^{*} ^(D), Yoon Joon Hwang, MD ^(D), Ji Young Lee, MD ^(D), Suk Hyun Bae, MD ^(D)

Department of Radiology, Ilsan Paik Hospital, Inje University College of Medicine, Goyang, Korea

Persistent trigeminal artery (PTA) represent an unusual remnant of the fetal carotid-basilar anastomosis. Persistent trigeminal artery variant (PTAV) is a rare anastomosis between the internal carotid artery and cerebellar artery, without an interposing basilar artery segment. We report the case of 49-year-old female with an incidentally discovered, rare variation of PTA that directly terminated in the ipsilateral superior cerebellar artery. The variation was observed on CT angiography, digital subtraction angiography, and MR angiography. Additionally, we reviewed the embryogenesis of PTA and PTAV and discussed the clinical implications of this variation.

Index terms Cerebral Arteries; Anatomic Variation; Angiography; Persistent Cerebral Embryonic Artery

INTRODUCTION

There are four major anastomoses that arise transiently between the carotid and vertebrobasilar systems: the primitive trigeminal, otic, hypoglossal, and proatlantal intersegmental arteries, named according to the cranial nerves they run parallel to (1). These vessels normally regress when final anastomoses are formed. Failure of regression of the trigeminal artery leads to a persistent carotid-basilar anastomosis (persistent trigeminal artery, PTA), which is the most frequent anastomosis found after birth, with an angiographic incidence of approximately 0.37% (2).

Persistent trigeminal artery variant (PTAV) originates from the precavernous portion of the internal carotid artery (ICA) and directly terminates at the cerebellar arteries without joining the basilar artery. It is extremely rare in general population. Previous Korean study reported the population-based incidence of PTAV approximately 0.32%



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*Corresponding author

Byung Hoon Lee, MD Department of Radiology, Ilsan Paik Hospital, Inje University College of Medicine, 170 Juhwa-ro, Ilsanseo-gu, Goyang 10380, Korea.

Tel 82-31-910-7694 Fax 82-31-910-7366 E-mail hoonbeer@hanmail.net

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ORCID iDs

Tae Young Park b https:// orcid.org/0000-0002-6022-6330 Byung Hoon Lee b https:// orcid.org/0000-0003-4481-7859 Yoon Joon Hwang b https:// orcid.org/0000-0003-3935-9208 Ji Young Lee b https:// orcid.org/0000-0002-6099-601X Suk Hyun Bae b https:// orcid.org/0000-0002-1888-2510

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on digital subtraction angiography (DSA) and 0.1% on MR angiography (MRA) (3). The most commonly reported PTAVs are the anterior inferior cerebellar artery (AICA) type (Saltzman type IIIb) (4, 5). Herein, we present a case of PTAV that directly terminated as the ipsilateral superior cerebellar artery (SCA) without joining the vertebrobasilar system that was incidentally detected on CT angiography (CTA), DSA, and MRA. To the best of our knowledge, the radiological findings of this condition have not yet been reported.

CASE REPORT

A previously healthy 49-year-old female was admitted to our emergency department with sudden onset of loss of consciousness after her neck was trapped between machines at her workplace for 5 minutes. Initial cervical spine CT revealed fractures of the left anterior tubercle of the C6 vertebra and the left transverse process of the C7 vertebra.

The patient subsequently underwent CTA of the head and neck, which demonstrated occlusion of the left proximal common carotid artery and reconstitution of the left carotid bulb via collaterals. Incidentally, a variation of PTA arising from the precavernous portion of the right ICA and running to the posterior fossa was identified. DSA of the right ICA revealed that this anomalous artery supplied the superior part of the right cerebellar hemisphere, which is the territory of the SCA. The anomalous artery did not connect with the basilar artery (Fig. 1A, B). Based on these findings, the patient was diagnosed with PTAV (Saltzman type IIIa) (5). No other vascular abnormalities, such as arteriovenous malformations and intracranial aneurysms, were observed.

The patient was managed conservatively and discharged 74 days after the trauma without focal neurologic deficit. A follow-up MRA at 15 months after the injury confirmed that the PTAV contributed to the ipsilateral SCA without joining the basilar artery (Fig. 1C-E).

This study has got an ethical review exemption from the Institutional Review Board of our institution and we achieved the written informed consent from the patient (IRB No. 2020-09-034).

DISCUSSION

Embryologically, four fetal carotid-basilar anastomoses temporarily exist: the primitive trigeminal, otic, hypoglossal, and proatlantal intersegmental arteries, named according to the cranial nerves they run parallel to (1). Generally, these fetal anastomoses regress during the formation of the posterior communicating artery. However, failure of their regression results in their persistence into adulthood. PTA is the most cephalad and most common variation among these anastomoses. During embryogenesis, incomplete fusion of the longitudinal neural arteries may lead to an absence of a direct connection between the PTA and basilar artery, resulting in its termination as one of the cerebellar arteries; this is known as PTAV (6).

In 1959, Saltzman (5) described the angiographic appearance of PTA and classified it into three types according to angiographic appearance. The Saltzman type I PTA connects to the basilar artery at a level between the SCA and AICA. The Saltzman type II PTA connects to the basilar artery above the origin of the SCAs. Saltzman type III PTA is characterized by the presence of a PTA arising from the precavernous ICA and connecting directly with a cerebel-

Unusual Persistent Trigeminal Artery Variant



Fig. 1. A right persistent trigeminal artery variant discovered incidentally in a 49-year-old female with cervical trauma.

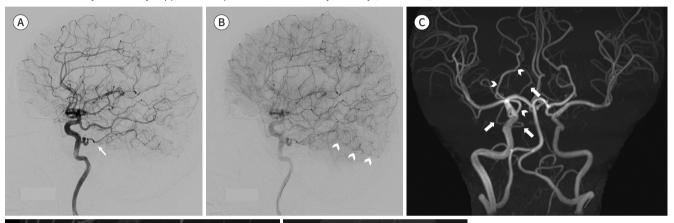
A. Lateral projection digital subtraction angiography image of the right internal carotid artery. Early arterial phase image depicts the persistent trigeminal artery variant (arrow), arising from the precavernous portion of the right internal carotid artery and running to the posterior fossa.

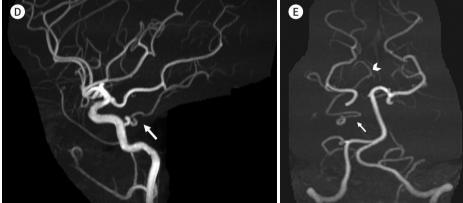
B. Late arterial phase image depicts that the persistent trigeminal artery variant supplies the superior part of the right cerebellar hemisphere (arrowheads).

C. Time-of-flight MR angiography maximum intensity projection image. The apicoposterior view of the intracranial arteries depicts the persistent trigeminal artery variant (arrows) running to the right superior cerebellar artery territory, below the right posterior cerebral artery (arrow-heads).

D. Lateral view of the right internal carotid artery depicts the persistent trigeminal artery variant (arrow) arising from the precavernous portion of the right internal carotid artery and running to the posterior fossa.

E. Apicoposterior view of the vertebrobasilar arteries depicts the persistent trigeminal artery variant (arrow), without an interposing segment of the basilar artery. This artery supplies the superior cerebellar artery territory (arrowhead).





lar artery without interposition of the basilar artery; it is also called PTAV. The PTAV arises from the ICA and terminates directly as the SCA (type IIIa), AICA (type IIIb), and posterior inferior cerebellar artery (type IIIc) without interposition of the basilar artery (Fig. 1F) (7). The present case was consistent with Saltzman type IIIa PTAV.

Generally, PTA and PTAV have been noted incidentally. However, the presence of an embryonic vessel persisting into adulthood indicates disturbed cerebrovascular development. Therefore, PTAV is frequently associated with other vascular abnormalities, particularly aneurysms (26%) (8). Common aneurysm sites associated with PTAV are the circle of Willis and the ICA-PTAV junction (8). Other vascular anomalies associated with PTA and PTAV are arteriovenous malformation (4.5%), carotid and vertebral artery agenesis, carotico-cavernous fis-

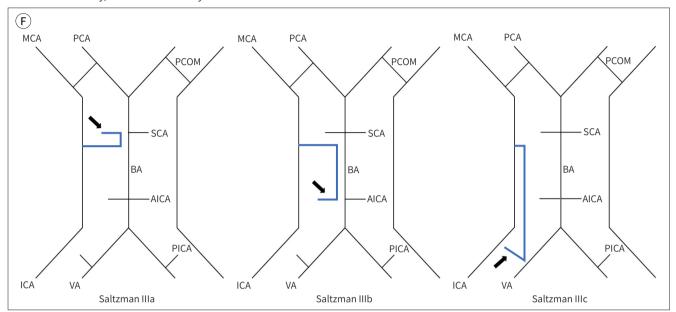
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Fig. 1. A right persistent trigeminal artery variant discovered incidentally in a 49-year-old female with cervical trauma.

F. Schematic illustration of Saltzman Type IIIa, IIIb, and IIIc PTA.

Focal non-fusion of the longitudinal neural artery with the trigeminal artery at the levels of the SCA, AICA, and PICA resulted in the PTA terminating in the SCA (Saltzman IIIa), AICA (Saltzman IIIb), and PICA (Saltzman IIIc), respectively (arrows).

AICA = anterior inferior cerebellar artery, BA = basilar artery, ICA = internal carotid artery, MCA = middle cerebral artery, PCA = posterior cerebral artery, PCOM = posterior communicating artery, PICA = posterior inferior cerebellar artery, PTA = persistent trigeminal artery, SCA = superior cerebellar artery, VA = vertebral artery



Modified from Ali et al. Neurosurgery 2008;62:E746-E748 (7).

tula, and Moyamoya disease (7-9). Therefore, neuroradiologists, neurologists, and neurosurgeons should be aware that PTAV could be accompanied by other vascular anomalies. In our case of PTAV, however, aneurysms or other vascular malformations were not detected.

It is vital to recognize these vessels before surgical treatment and endovascular intervention. Because blood flows from the carotid to the vertebrobasilar system, there is a risk of brainstem or cerebellar ischemia caused by emboli from an ulcerated atherosclerotic plaque in the carotid bulb passing through these anomalous vessels during an endovascular procedure (10).

In conclusion, we present an extremely rare case of PTAV, which directly terminates in the SCA (Saltzman type IIIa). To the best of our knowledge, the radiological features of this type of PTAV have not yet been reported. Although PTA and PTAV are rare, careful investigation of these anastomoses is important. Inappropriate diagnosis could lead to serious complications, such as hemorrhage or brainstem and cerebellar ischemia, during interventional endovascular procedures or surgeries.

Author Contributions

Conceptualization, L.B.H.; data curation, B.S.H.; investigation, L.J.Y.; methodology, B.S.H.; project administration, L.B.H., H.Y.J.; supervision, L.B.H.; validation, H.Y.J.; visualization, P.T.Y.; writing—original draft, P.T.Y.; and writing—review & editing, L.B.H.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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상소뇌동맥영역을 공급하는 지속성 원시 삼차신경동맥 변이

박태영·이병훈*·황윤준·이지영·배석현

지속성 원시 삼차신경동맥은 퇴화하지 않고 남아있는 내경동맥-기저추골동맥 문합이다. 변 형 지속성 원시 삼차신경동맥은 드문 비정상적인 문합으로, 내경동맥과 기저동맥의 연결 없 이 내경동맥에서 직접 혈관이 분지 되어 소뇌 동맥의 영역에 혈액을 공급한다. 본 연구진들 은 그중에서도 변형 지속성 원시 삼차신경동맥이 동측의 상소뇌동맥 영역에 직접 혈액을 공 급하는 특히 드문 증례를 발견하였다. 시행된 전산화단층촬영 혈관조영술, 디지털감산 혈관 조영술, 자기공명 혈관조영술 사진을 이용하여 이를 보고하고자 한다. 추가적으로 본 연구진 들은 지속성 원시 삼차신경동맥과 변형 지속성 원시 삼차신경동맥의 발생학과 임상적 의미 에 대하여 검토하였다.

인제대학교 의과대학 일산백병원 영상의학과