

Ascending pharyngeal artery supplying the posterior inferior cerebellar artery via the hypoglossal canal with preserved anastomosis to the vertebral artery: a rare variant of the persistent hypoglossal artery

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SUMMARY

Carotid-vertebrobasilar anastomoses are rare and usually discovered serendipitously; the persistent hypoglossal artery is the second most common, with an incidence of 0.29%. We present a very rare anatomical variant of a persistent hypoglossal artery. This variant was visualized on 2D and 3D angiography and on MRI.

In the case reported here, the hypoglossal branch of the neuromeningeal trunk of the ascending pharyngeal artery communicated with the posterior inferior cerebellar artery (“type 2 persistent hypoglossal artery variant” of Uchino’s classification), but with a preserved junction with the vertebral artery, which is not hypoplastic (“ascending pharyngeal artery - vertebral artery anastomosis” of Lasjaunias’ description). A review of the literature was performed on the “type 1-2 persistent hypoglossal artery variant” (11 cases) and schematic representations of the

different anatomic variants are illustrated. The pharyngo-cerebellar artery and pharyngo-vertebral anastomosis are variants of the persistent hypoglossal artery, these relatively small branches may be missed, with a risk of cerebellar infarction during embolization.

Key words: Cerebellar arteries – External carotid artery – Vertebrobasilar system – Radiological anatomy – Interventional neuroradiology

INTRODUCTION

Carotid-vertebrobasilar (VB) anastomoses are rare and usually discovered serendipitously. However, they may have clinical significance, including aneurysm related to the anomalous artery (Tse et al., 2019), cerebral ischaemia owing to single arterial supply, and dangerous anastomoses in the case of embolization. Four types of

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foetal anastomoses exist between the carotid artery and the VB system at 5 weeks gestation (at the 3-5 mm stage) (Padget, 1948); from the caudal to the cranial position: proatlantal intersegmental, hypoglossal, otic, and trigeminal arteries. During development, these primitive arteries regress, and/or fuse, to form the mature arterial system and with the development of the posterior communicating (PcomA) and vertebral (VA) arteries. Nevertheless, they can persist in some cases. The persistent trigeminal artery (PTA) is the most common persistent primitive carotid-VB anastomosis (Ota and Komiyama, 2022). Each persisting primitive artery can lead to several anatomical variations.

The persistent hypoglossal artery (PHA) is the second most common persistent carotid-VB anastomosis, with an incidence ranging from 0.027% (Lie and Foundation, 1968) in historic angiographic studies to 0.29% (Uchino et al., 2013) in a recent study based on 2074 CT angiographic images. Many cases have been reported since Batujeff (Batujeff, 1889) first reported in 1889 PHA in an autopsy case, and Begg (Begg, 1961) by angiography in 1961. Based on the work of Brismar (Brismar, 1976), three essential imaging diagnostic criteria are required to describe a PHA: 1) the PHA leaves the proximal ICA as an extracranial branch; 2) the PHA ascends posterior to the cervical ICA and passes into the posterior fossa via the hypoglossal canal; 3) the PHA joins the V4 segment of the ipsilateral VA or the caudal portion of the basilar artery (BA).

According to this description, a PHA is usually large and associated with an hypoplastic ipsilateral VA. Bilateral PHAs are rare (Murayama et al., 1985).

PHAs from the external carotid artery (ECA) have been reported recently. This extremely rare artery arises from the proximal ECA, and its proximal segment rises anterior to the cervical ICA. To our knowledge, 12 cases have been published (Welten et al., 1988; Nakamura et al., 2000; Meguro et al., 2007; Lee et al., 2010; Uchino and Saito, 2011; Nanto et al., 2012; Uchino et al., 2013; He et al., 2014; Sabouri et al., 2014; Yamamoto et al., 2019) (the homolateral VA was hypoplastic or absent in all cases). Based on this anatomical

observation, Uchino proposed naming the usual PHA with an ICA origin as follows: “type 1 PHA”; and PHA with an ECA origin: “type 2 PHA” (Uchino and Saito, 2011). As proposed by Anderson and Sondheimer (Anderson and Sondheimer, 1976), entry into the hypoglossal canal is the discriminating factor differentiating the PHA from the persistent proatlantal artery (joining the VA via the foramen magnum) or the PTA (joining the BA via the prepontine cistern). In adults, the remnant of the PHA is the hypoglossal branch of the ascending pharyngeal artery (APA) (Lasjaunias et al., 1981, 2013). The APA normally arises from the posterior wall of the proximal ECA; after a short common trunk, the APA divides into the pharyngeal trunk anteriorly, the neuromeningeal trunk posteriorly and the inferior tympanic artery. The neuromeningeal trunk is intracranial, dividing into hypoglossal and jugular branches, entering the posterior fossa through the hypoglossal canal and the jugular foramen, respectively. The hypoglossal branch supplies the vasa nervorum of the hypoglossal nerve and the meninges of the posterior cranial fossa where it is in balance with the other arteries of this region (Hacein-Bey et al., 2002).

Among type 1-2 PHAs, a variant (“type 1-2 PHA variant”) may exist as a small PHA supplying only the posterior inferior cerebellar artery (PICA) without connecting to the BA. Similarly, for the “PTA variant”, the cerebellar artery originates from the PTA (anterior inferior cerebellar artery [AICA], superior cerebellar artery [SCA] and/or PICA) without persistent connection to the BA (Uchino, 2019).

We report here a very rare anatomical variation: a type 2 PHA variant with a persisting PICA-VA junction and without hypoplastic VA.

CASE REPORT

This anatomical variant was serendipitously discovered in our centre during a right lateral sinus stenting procedure for idiopathic intracranial hypertension syndrome with disabling pulsatile tinnitus caused by lateral sinus stenosis. Images were obtained from a preoperative brain MRI with 3D TOF sequence and cerebral arteriography per-

formed via the right radial approach during the stenting procedure with selective catheterisation of the right common carotid and vertebral arteries. The Institutional Review Board (IRB) committee approved the study. The procedures were carried out after obtaining an informed consent and in accordance with ethical standards.

In the case reported here, angiograms of the common carotid and vertebral arteries showed:

1) the VAs were co-dominant and of normal calibre; 2) the APA branched off as expected from the ECA and supplied the pharyngeal arteries; 3) the hypoglossal branch of the neuromeningeal trunk of the right APA was voluminous and fed the PICA through the right hypoglossal canal; 4) there was preserved hypoplastic communication between the VA and the PICA, with the connection occurring shortly after the exit of the hypoglossal canal,

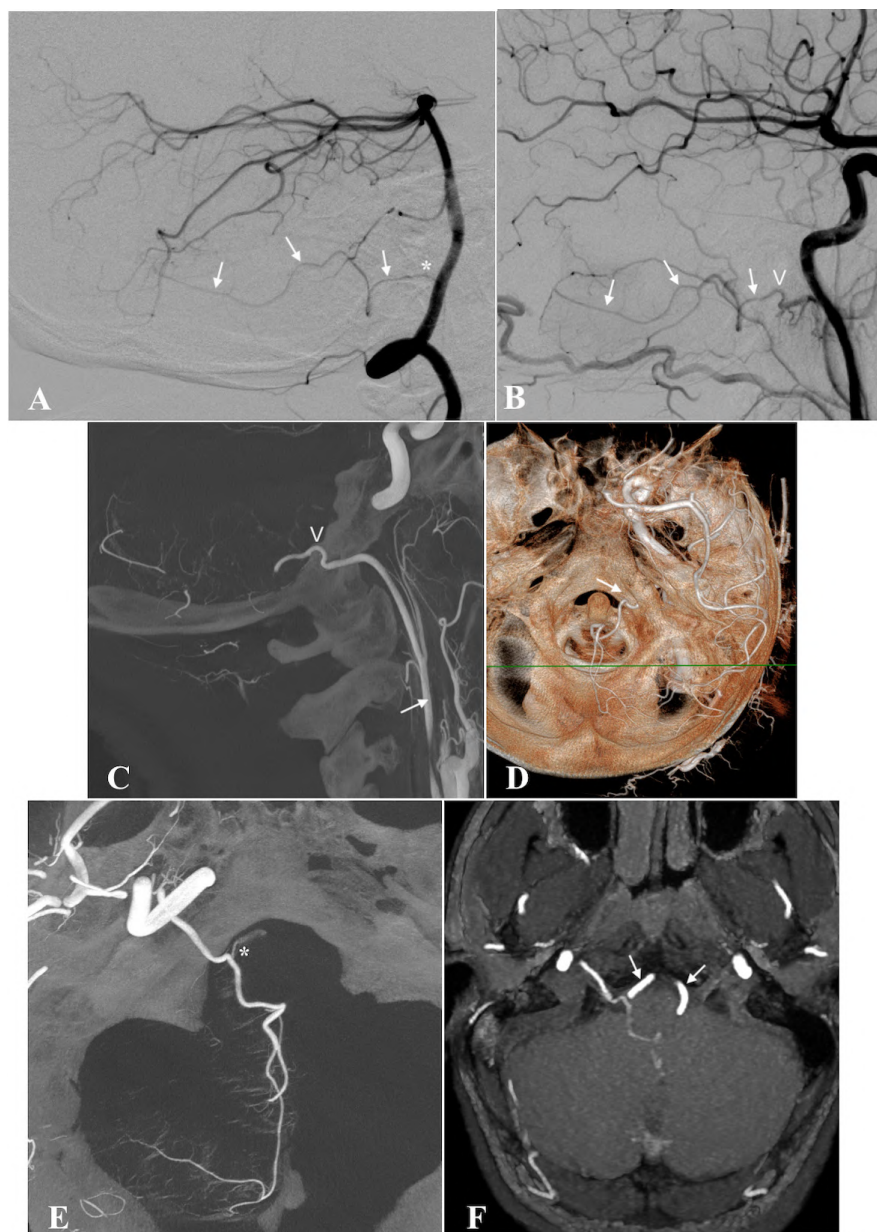


Fig. 1.- Case report illustration. **A:** Selective angiography of the right VA, lateral view, showing normal caliber of the V4 segment and persistence of a narrow communication (asterisk) with the right PICA (arrows); **B:** Selective angiography of the right CCA, lateral view, demonstrating the APA (hypoglossal) - PICA anastomosis (arrowhead); **C:** 3D rotational DSA, lateral view, finding the APA (arrow) originating from the external carotid artery with the pharyngeal territory located anteriorly and posteriorly, through its hypoglossal branch (arrowhead), the neuromeningeal branch takes over the territory of the homolateral PICA; **D:** 3D-DSA volume rendering showing the emergence of the APA-PICA at the hypoglossal foramen (arrow); **E:** 3D-DSA with MIP, the persistent communication between PICA and V4 is visible (asterisk), **F:** 3D TOF MRI showing codominance and non-hypoplastic V4 segments on both sides (arrows), the V4-PICA communication is not visible. VA: vertebral artery, PICA: posterior-inferior cerebellar artery, CCA: common carotid artery, APA: ascending pharyngeal artery, DSA: digital subtraction angiography, MIP: maximum intensity projection, TOF: time-of-flight.

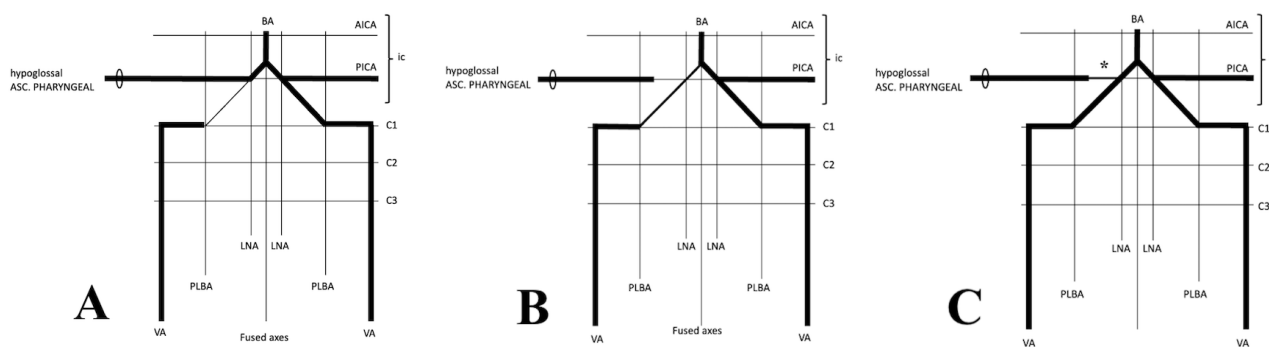


Fig. 2.- Frontal schematic representation of hypoglossal variations on the vertebrobasilar system. **A:** PHA; **B:** APA origin of PICA (pharyngo-cerebellar variety); **C:** Patient of this case report, APA (hypoglossal) – VA anastomosis with persistent of a hypoplastic stem of PICA (asterisk). On the three diagrams, the left PICA is represented supplied by the V4 segment (most frequent course, as in our patient). PHA: persistent hypoglossal artery, VA: vertebral artery, PICA: posterior-inferior cerebellar artery, APA: ascending pharyngeal artery, LNA: longitudinal neural artery, PLBA: primitive lateral basilovertebral artery, BA: basilar artery, AICA: antero-inferior cerebellar artery, ic: intracranial.

there was no accessory PICA visible on the VA; 5) the APA terminated in the PICA and hemodynamically supplied the entire PICA territory, the supply of blood flow into the PICA by the VA was accessory (Fig. 1). This anatomical pattern represents an intermediate form between the type 2 PHA and the type 2 PHA variant, where there is no communication with the VB system (Fig. 2).

DISCUSSION

The hypoglossal canal is formed by the fusion of the intervertebral foramen of the area vertebralis (Karasu et al., 2009). The hypoglossal canal contains three important structures (Karasu et al., 2009): the twelfth cranial nerve, a meningeal branch of the APA, and a surrounding emissary venous plexus that communicates between the basilar venous plexus and the marginal sinus

Table 1. Type 1-2 PHA variant in the literature.

N°	Author	Year	Sex	PHA Variant	Side	Imaging	Homolateral VA	Associated arterial anomalies
1	Teal JS	1973	M	1	R	DSA	Hypoplastic	Ruptured AComA aneurysm, infraoptic course of the right ACA
2	Lasjaunias P	1981		1	R	DSA	Hypoplastic	Bilateral PHA
3	Murayama Y	1985	M	1	R	DSA	Hypoplastic	Bilateral PHA, ruptured PHA-BA aneurysm, right MCA aneurysm
4	Andoh K	2001	F	1	L	DSA, CTA	Hypoplastic	Right VA fusiform aneurysm, left MCA saccular aneurysm
5	Nakanishi N	2004	M	1	R	MRA, DSA	Absent	No
6	Kim JT	2009	F	2	L	MRA, CTA, DSA	Present	Occlusion of the left MCA with rich collateral supplies, severe stenosis of the right proximal MCA (Moya-Moya?)
7	Uchino A	2013	M	2	R	MRA, CTA	Present	Narrowing of entire right ICA
8			F	2	L	MRA, CTA	Absent	Right MCA aneurysm
9	Namba K	2017		2	R	DSA		
10	Patira R	2017	M	1	L	CTA	Absent	Bilateral PHA
11	Uchino A	2018	F	1	R	MRA	Absent	No

PHA: persistent hypoglossal artery, **VA:** vertebral artery, **BA:** basilar artery, **MCA:** middle cerebral artery, **ACA:** anterior cerebral artery, **AComA:** anterior communicating artery, **ICA:** internal carotid artery, **DSA:** digital subtraction angiography, **CTA:** computed tomography angiography, **MRA:** magnetic resonance angiography, **R:** right, **L:** left, **M:** male, **F:** female

around the foramen magnum. The hypoglossal artery and nerve are related to the rhombomere 8 and occipital somites 2-4 (Müller and O’Rahilly, 2011). The hypoglossal nerve is phylogenetically equivalent to a spinal nerve and not to a cranial nerve, although, given its evolutionary history, it is associated with the head into which it is secondarily included (Louryan and Vanmuylder, 2018).

To our knowledge, there are reports of 7 cases of type 1 PHA variant (Teal et al., 1973; Murayama et al., 1985; Andoh et al., 2001; Nakanishi et al., 2004; Uchino and Suzuki, 2018) and 4 cases of type 2 PHA variant (pharyngo-cerebellar artery) (Lasjaunias et al., 1981, 2013; Kim et al., 2009; Uchino et al., 2013) (Table 1). Most often, they are located on the right side (7/11) and the homolateral VA is hypoplastic or absent.

Lasjaunias et al. (1981, 2013) assume that the remnant of the PHA in adults is the hypoglossal branch of the neuromeningeal trunk of the APA. This hypothesis may explain the presence of a type 2 PHA. Normally, the hypoglossal branch of the APA supplies the meninges of the posterior fossa, the vasa nervorum of the hypoglossal nerve (XII), and a posterior descending branch contributes to the odontoid arch system providing anastomoses to the VA (Hacein-Bey et al., 2002). Whereas the lower cranial nerves are supplied by the VB system, the supply to their foraminal parts is mainly ensured by the neuromeningeal trunk of the APA (Lasjaunias et al., 2013). The dural branches of the jugular branch of the APA also rarely anastomose with the PICA through the jugular foramen (Effendi et al., 2016), although this does not represent a type of PHA variant (Uchino, 2019). According to Morris and Moffat (Moffat and Morris, 1956), the PHA is not identical to the embryonic hypoglossal artery. They suggest that the PHA is composite and is composed of three parts: 1) the primitive hypoglossal artery, 2) portions of the primitive lateral basilovertbral anastomosis (PLBA) (lateral anastomotic channel), and 3) the transverse anastomotic channels connecting the PLBA to the longitudinal neural artery (LNA). The type 1-2 PHA variant results from the persistence of the first and second parts, from involution or failure to develop of the third part and from disconnection of the PICA origin from the VA (Andoh et al., 2001).

Lasjaunias et al. (2013) theorize the existence of an “APA-vertebral anastomosis” with homolateral non-hypoplastic V4 segment of the VA. An illustration of this situation, similar to that for our patient, is provided on page 216 of the book *Surgical Neuroangiography* (Lasjaunias et al., 2013). This, to our knowledge, is the only other case. Recently, Bordes et al. (2021) report a case of hypoplastic pharyngo-vertebral anastomosis without homolateral V4 hypoplasia, but with PICA branched off the VA. This rare variant could be explained by a minimal persistence of the third portion of the PHA, or by the persistence of the origin of the PICA on the VA that is associated with a patent homolateral V4 segment of the VA. It is possible that the spatial resolution of MRA and CTA is insufficient to visualise a persistent hypoplastic communication with the VA (as in our case) and that, for some of the cases of type 1-2 PHA variant described in the literature where DSA is not performed, there is in fact a patent PICA-VA junction.

Unlike PHA, which is usually large, the type 1-2 PHA variant is a relatively small branch and may be missed, with a risk of cerebellar infarction during embolization. Thus, the existence of a type 1 PHA variant should not be ignored before planning a temporary intraoperative carotid occlusion for carotid endarterectomy, endovascular carotid occlusion, or balloon carotid artery occlusion testing. Recognition of a type 2 PHA variant is very important for endovascular treatment of a dural arteriovenous fistula of the hypoglossal canal/sigmoid sinus/jugular gulf fed by the APA, for preoperative embolization of a petroclival meningioma, and for preoperative or haemostatic embolization of pharyngeal tumours. The persistence of a PICA-VA junction could minimize these risks, provided that the flow via the VA is sufficient to compensate for the PICA territory.

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