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CORRESPONDENCE

Bilateral Persistent Trigeminal Arteries, One of Them Ending in the Posterior Inferior Cerebellar Artery

Case Report and Review of the Literature

D. Meila · K. Papke · M. Schlunz-Hendann · A. Mangold · C. Jacobs · F. Brassel

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Introduction

The persistent trigeminal artery (PTA) is the most common embryologic anastomosis between the carotid and vertebrobasilar systems. It was found in 0.1–0.6% in large angiographic series [1]. Other persistent embryonic intracranial and extracranial vessels are the hypoglossal and proatlantal arteries. The otic artery, another reported embryologic anastomosis, is a highly controversial artery and presumably does not exist [2]. PTA variants, defined as arteries with direct anastomosis between the intracranial internal carotid artery and the cerebellar arteries, without the interposition of the basilar artery, were found in 0.18% of angiographic studies [3]. However, bilateral PTA is a very rare condition [4, 5]. The case of a patient with bilateral PTA, one of them ending in the posterior inferior cerebellar artery (PICA) is presented. To our knowledge this is the first case reported in the literature with such an anatomic condition. The embryological origin, the Saltzman's classification and the clinical significance of this uncommon anatomic variation are discussed.

Dr. med. D. Meila (✉) · Dr. med. M. Schlunz-Hendann · Dr. med. A. Mangold · Prof. Dr. med. F. Brassel
Department of Radiology and Neuroradiology,
Klinikum Duisburg, Zu den Rehwiesen 9,
47055 Duisburg, Germany
e-mail: dmeila@yahoo.de

Dr. med. K. Papke
Department of Radiology, St. Bonifatius-Hospital,
Lingen, Germany

Dr. med. C. Jacobs
Department of Orthodontics, University of Mainz,
Mainz, Germany

Case Report

A 24-year-old-woman was transferred to the emergency department with oculomotor cranial nerve (CN) III palsy on the right side 2 days after a caesarean section. The clinical neurological examination showed ophthalmoplegia, ptosis and mydriasis as well as diplopia which was worse in left gaze. No other neurological deficit was found. On presentation computed tomography angiography (CTA) was immediately performed and revealed a 7 mm right-sided internal carotid artery-posterior communicating artery (ICA-Pcom) aneurysm. The patient underwent a cerebral angiogram which confirmed the diagnosis. Incidentally, on the same side of the aneurysm a PTA ending directly in the PICA territory without opacification of the basilar artery was noted (Figs. 1, 2a). Injection of the right vertebral artery showed the lack of the PICA territory (Fig. 2b). Selective angiography of the left ICA revealed another PTA, with filling of the vertebro-basilar system, leading to the coincidental diagnosis of a bilateral PTA (Figs. 3a, b). Both PTAs were of the lateral type, running laterally to the dorsum sellae. The patient was treated with platinum detachable coils (Fig. 2a). ICA-Pcom aneurysm-induced CN III palsy improved after complete coil embolization.

Discussion

In the early development of the embryo three longitudinal artery systems exist bilaterally, two ventral systems correspond to the ventral and dorsal aorta while the third, the dorsal one, is located on the midline. The dorsal system corresponds to the paired longitudinal neural arteries. The trigeminal artery develops in the embryo at the 4 mm stage and bridges the distal dorsal aorta with the paired longitu-

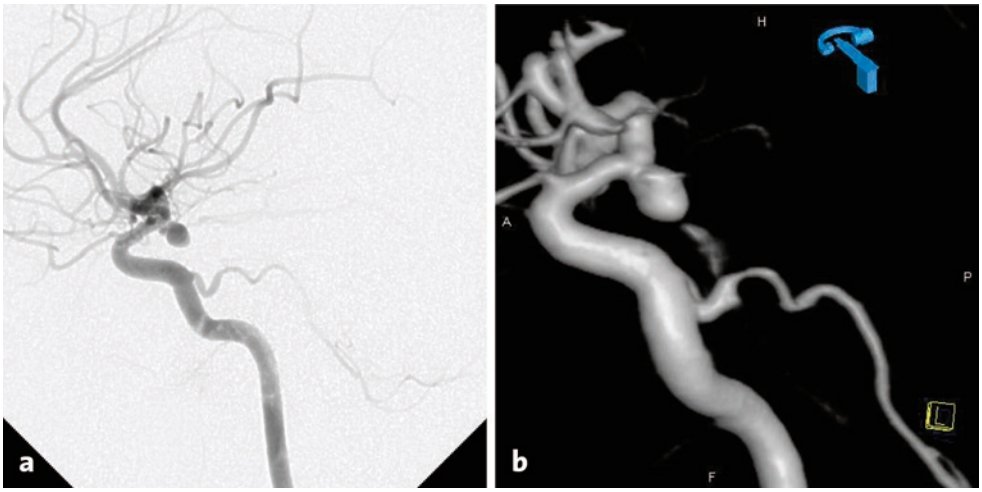

Fig. 1 **a, b** Right internal carotid artery injection in lateral projection and 3D rotational angiographic reconstruction, demonstrating the ICA-Pcom aneurysm and the PTA directly feeding the PICA

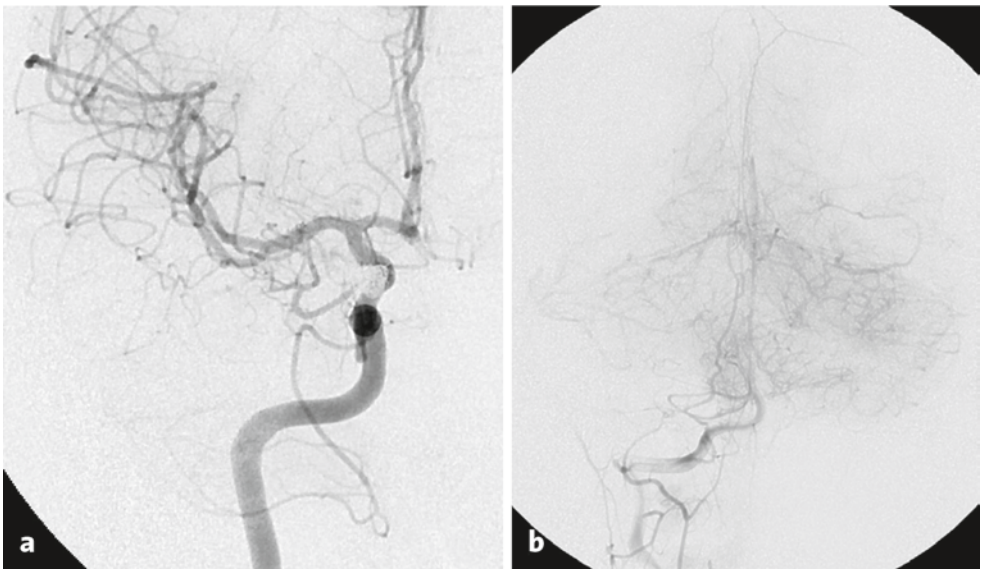

Fig. 2 **a** Right internal carotid artery injection in frontal projection showing PICA filling on the right and coil package after ICA-Pcom aneurysm embolization. **b** Townes projection of a right vertebral artery injection in the early capillary phase demonstrating the lack of the PICA territory

dinal neural artery system. Intersegmental longitudinal anastomoses develop caudally to the trigeminal artery (Fig. 4a). These new channels form the later vertebral arteries at the end of the second phase, according to Padget [6]. At the 7–12 mm stage the trigeminal artery regresses. The basilar artery is finally the result of two simultaneous fusions: the caudal division of the internal carotid arteries cranially to the trigeminal artery and caudally the fusion of the paired longitudinal neural arteries [7] (Fig. 4b).

The persistent trigeminal artery (PTA) is the most common persistent embryologic anastomosis between the carotid and vertebro-basilar systems. It was found in 0.1–0.6% in large angiographic series [1]. Bilateral PTA is a very rare condition and only six cases were reported in the literature but six other described cases of a PTA variant terminating as a PICA were available to be reviewed systematically in the literature (Table 1).

Quain first described the anatomy and Sutton published the first angiographic observation of the PTA [18, 19]. The PTA originates from the ICA at the point where the vessel leaves the carotid canal and penetrates the cavernous sinus. The artery passes the latter medial to the ophthalmic branch of the trigeminal nerve via Meckel's cave and reaches the posterior fossa. The PTA may either run laterally to the dorsum sellae or have a middle course through or over the dorsum sellae to communicate with the basilar artery [20].

Saltzman [21] was the first to study the trigeminal arteries by cerebral angiography. In a recent paper Ali et al. [16] reviewed the Saltzman's classification. In Saltzman type 1 the PTA supplies mainly the distal basilar artery, the superior cerebellar arteries (SCA) and the posterior cerebral arteries (PCA). In Saltzman type 2 the PTA joins the basilar artery above the origin of the SCAs and the PCAs receive the blood supply through patent posterior communicating arteries (Pcom). Saltzman type 3 is considered to be a combination of types 1 and 2, with the anastomosis supplying the SCAs bilaterally as well as the contralateral PCA. Variants of the PTA, in which there is no interposition of the

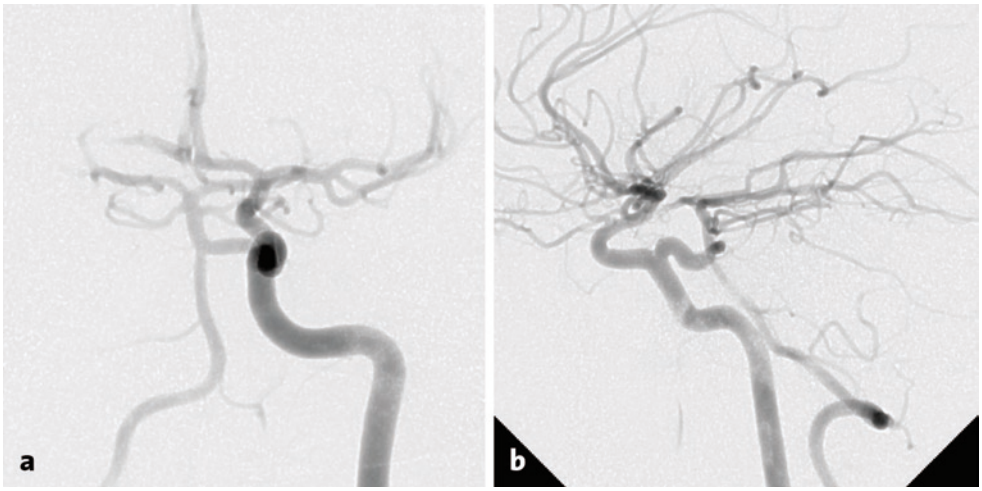

Fig. 3 Left internal carotid artery injection in the frontal (**a**) and lateral (**b**) projection demonstrating PTA with opacification of the vertebro-basilar system

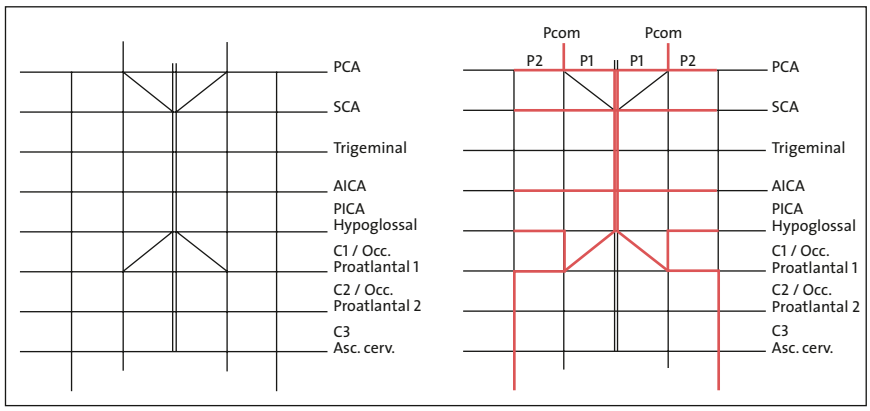

Fig. 4 **a** Matrix for the vertebro-basilar system according to the persistent embryonic arteries. **b** Matrix showing the normal schematic vertebro-basilar system without any persistent embryonic vessel. (Modified from [7])

basilar artery were classified by Ali et al. as Saltzman type 3a with a PTA terminating in the SCA, Saltzman type 3b with a PTA ending in the AICA and in Saltzman type 3c with a PTA ending directly in the PICA.

In a recent work O'uchi and O'uchi [22] analyzed 103 cases of PTAs. They emphasized that from an embryological point of view supratentorial and infratentorial arteries are totally different. The PCA or the posterior communicating artery and the PTA have no developmental relationship to each other. As a result, they stated that the original Saltzman's classification of the PTA has no meaning. However, the revised Saltzman's classification by Ali et al. [16] facilitates the description of the PTA variants without communication with the basilar artery. The PTA can take different courses of the longitudinal neural arteries, even without communication with the basilar artery. This was well demonstrated by Ali and colleagues for the Saltzman types 3a–c. The case described here shows on the left a Saltzman type 1 with a PTA mainly supplying the distal basilar artery, the superior cerebellar arteries (SCA) and the posterior cerebral arteries (PCA). On the right side a Saltzman type 3c is demonstrated with a right PTA ending directly in the PICA (Fig. 5) and shows the schematic application of the reviewed Saltzman's classification. The matrix for the vertebro-basilar system according to the persistent embryonic arteries (Fig. 4a) facilitates the recognition and understanding of arterial variations in clinical practice additionally.

O'uchi and O'uchi [22] divided the PTAs into a lateral and a medial type, according to their course. The PTA may run lateral to the dorsum sellae or have a middle course through or over the dorsum sellae. In the case described here both PTAs were of the lateral type.

To our knowledge, this is the first case of bilateral PTA, one of them supplying directly the PICA territory, with no basilar artery opacification (Fig. 5)

Arterial variations, as well as associated intracranial aneurysms or other cerebrovascular diseases, e.g. carotid-cavernous fistula, were reported to be associated to embryological persistent arteries, in particular to the PTA [23, 16, 24–26]. PTA and its variants have often been found incidentally. Some authors reported the prevalence of PTA with associated intracranial aneurysms to be 14–32% [27, 28]. According to Cloft et al. this prevalence range was unreliable due to collections of published reports. In their own unbiased study they found the prevalence of intracranial

Table 1 Cases with similar embryological anastomoses and associated anomalies

Reference	Age, Sex	Anastomosis	Symptoms/diagnosis	Associated anomalies
Binet and Young [8]	NA	Bilateral PTA	NA	Right ICA hypoplasia, left ICA aneurysm
Chambers and Lukin [9]	59, F	PTA ending in PICA	Incidental	NA
Binet and Young [8]	60, F	Bilateral PTA	Headache, gait disturbance, cerebellar tumor	NA
Taguchi et al. [10]	45, F	Bilateral PTA	Diplopia, exophthalmos	NA
Manabe et al. [11]	38, F	PTA ending in PICA	Subarachnoid hemorrhage	Multiple cerebral aneurysms
Okada et al. [12]	43, M	Bilateral PTA	Hemiparesis, brain stem infarct	Bilaterally absent vertebral artery at origin
Chen and Liu [13]	64, F	Bilateral PTA	Severe headache, vomiting, intraventricular hemorrhage	Moyamoya disease
Hui et al. [14]	37, M	PTA ending in PICA	Headaches, proptosis, chemosis	Ipsilateral dural cavernous AVM
Nishio et al. [15]	69, F	PTA ending in PICA	Diplopia, ophthalmoplegia	Ipsilateral non-ruptured cavernous ICA aneurysm
Ali and Walker [5]	55, F	Bilateral PTA	Incidental	Bilateral internal carotid aneurysms
Ali et al. [16]	66, F	PTA ending in PICA	Left pulsatile tinnitus, proptosis, conjunctival injection, diplopia and decreased visual acuity	Ipsilateral carotid-cavernous fistula
Raphaeli et al. [17]	53, F	PTA ending in PICA	Global aphasia, hemiparesis, intracerebral hematoma	Cerebral aneurysm

AVM arteriovenous malformation, *ICA* internal carotid artery, *F* female, *M* male, *NA* not available, *PICA* posterior inferior cerebellar artery, *PTA* persistent trigeminal artery

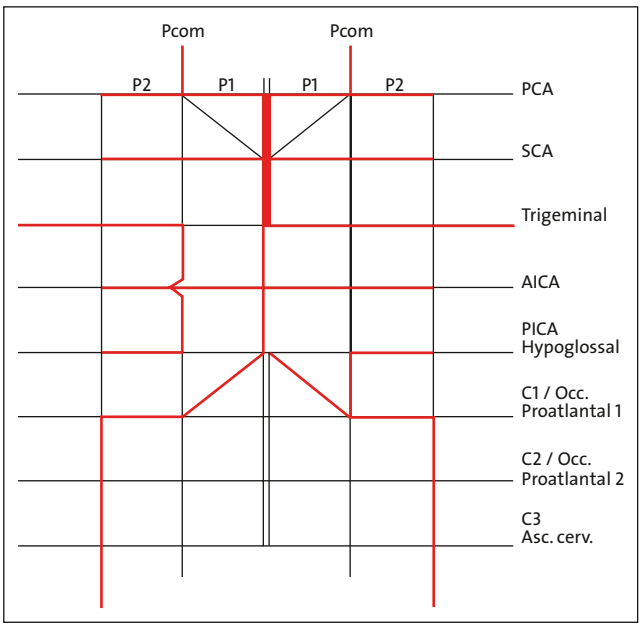

Fig. 5 Present case with bilateral PTA is demonstrated. Saltzman type 1 showing a left trigeminal artery (PTA) with main supply to the distal basilar artery, the superior cerebellar arteries (SCA) and the posterior cerebral arteries (PCA) and Saltzman type 3c is demonstrated with a right PTA ending directly in the PICA

aneurysms in patients with PTA to be not greater than in the general population [29].

Another vascular anomaly, concerning a vertebral artery ending in PICA, has been described in association with moyamoya disease [30]. Bilateral PTA, even a very rare anatomic finding, was also shown to be associated to intracranial aneurysms [4, 5]. We add this case with this rare anatomic condition and an associated cerebral aneurysm.

The clinical significance of the PTA and its variations is amongst others documented in two cases with brain stem infarction and bilateral occipital infarctions [12, 31]. Our case shows the supply of the PICA-territory from the anterior circulation. In a case of an ischemic event in the PICA-territory and a coexistent ICA-stenosis on the same side further diagnostic investigations should be performed to confirm a possible PTA.

The findings of the PTA and its associated variations may be incidentally, but its role in endovascular procedures like trapping, stenting, coiling, other embolization procedures or neurosurgical techniques are essential due to the rare, uncommon anatomic condition. Knowledge and understanding of embryologic anatomy is important in therapeutic decisions and can avoid hemorrhagic or ischemic complications.

Conflict of Interest Statement The authors declare that there is no potential conflict of interest in relation to this article.

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