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ORIGINAL ARTICLE

Persistent trigeminal artery associated with an occipital arteriovenous malformation: a case report and literature review

Luis Alberto Rodriguez-Hernandez[1] · Pablo Martinez[2] · Iván Baracaldo[2] · Monica Alonso[3] · Diego Lopez-Mena[2] · Héctor Alfredo Montenegro Rosales[2]

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Abstract

Background Persistent trigeminal artery or persistent primitive trigeminal artery (PTA) is the most common carotid-basilar anastomosis in both cadaveric and live patient studies, followed by persistent hypoglossal and persistent otic arteries. Approximately 0.2% of all angiographies reported this finding.

Case report: We present the case of a 21-year-old male who arrived at the emergency department with tonic–clonic seizures. After performing diagnostic contrast magnetic resonance imaging and digital subtraction angiography, the patient was diagnosed with a right occipital arteriovenous malformations (AVM) fed by the right calcarine artery associated with an ipsilateral PTA. After considering surgical and endovascular treatment options, the patient was selected for watchful waiting. We included a literature review of the PTA, the results of a PubMed search regarding the combined presence of these findings, and a brief discussion providing insight into the implications for treatment.

Conclusions Although several studies have linked PTA to different vascular pathologies, such as cerebral aneurysms, the association between PTA and AVMs remains scarce. This case, along with the literature review, shows that further research is needed to characterize the relationship between these findings.

Keywords Persistent trigeminal artery · Arteriovenous malformation · Anatomic variations · Angiogram · Neuroradiology · Neurology

✉ Héctor Alfredo Montenegro Rosales
dr.montenegrorx@gmail.com

Luis Alberto Rodriguez-Hernandez
luis_albertorh@hotmail.com

Pablo Martinez
pablomartineza.98@gmail.com

Iván Baracaldo
ibaracaldo@innn.edu.mx

Monica Alonso
monik200655@hotmail.com

Diego Lopez-Mena
diegolmena@gmail.com

[1] Department of Neurosurgery, Instituto Nacional de Neurología y Neurocirugía, Mexico City, Mexico

[2] Department of Interventional Neuroradiology, Instituto Nacional de Neurología y Neurocirugía, Mexico City, Mexico

[3] Department of Neurology, Universidad del Sinu, Cordoba, Colombia

Introduction

The persistent trigeminal artery (PTA) is the most common carotid-basilar anastomosis. Eighty-five percent of the reported literature of carotid-basilar anastomosis concerns PTA [16]. PTA was described in live patients by Sutton in 1950 using cerebral angiography [2].

It is not uncommon for PTA to be associated with intracranial vascular pathology, especially intracranial aneurysms [15]. However, arteriovenous malformations (AVMs) remain a much less studied subject, with only a few cases reported. Herein, we present a case of a young male patient with a right occipital AVM associated with an ipsilateral PTA.

Case report

A 21-year-old male with a medical history of smoking and a remitted case of acute lymphoblastic leukemia was brought to the emergency department after presenting with a

generalized tonic−clonic seizure with loss of consciousness of unknown length. The patient denied any previous history of seizures or family history of epilepsy.

Initial magnetic resonance imaging (MRI) of the patient revealed a right posterior AVM (Fig. 1). The patient was started on antiepileptic medication and was scheduled for further testing.

Diagnostic subtraction angiography (DSA) demonstrated an occipital AVM fed by the calcarine artery measuring 11 mm in the dorsoventral axis and 17 mm in the lateral–lateral axis (Fig. 2b); it drained into the right occipital cortical vein and further into the superior sagittal sinus. The AVM was scored as grade II using the Spetzler–Martin classification (1 point for the nest size and 1 point for the eloquent area) and as grade I using the Buffalo classification (1 point for eloquence). DSA also revealed a persistent trigeminal artery type Saltzman 1 in the anterior and posterior projections (Fig. 2a).

At the moment that this article was written, the patient had adequate control of epileptic seizures and was selected for watchful waiting by consensus of the Interventional Neuroradiology and Vascular Neurosurgery departments.

Discussion

PTA is a carotid-basilar anastomosis that emerges in the early days of the embryologic development phase and disappears when the embryo reaches 11.5–14 mm [2]. It is the most common carotid-basilar anastomosis and is found in between 0.1 and 3% of the general population [7]. There are two kinds of the PTA: lateral (usual) type and medial (intrasellar) type. The latter one is rare.

As described by Saltzman, PTA might be classified as one of three types. In type 1, as in our case, PTA supplies most of the vertebrobasilar circulation, whereas type 2 PTA supplies only the superior cerebellar arteries. Type 3 usually refers to a PTA that ends in a cerebellar artery completely bypassing the basilar artery [9].

We performed a PubMed search using the following MESH terms: Persistent Trigeminal Artery, Persistent Primitive Trigeminal Artery, and Arteriovenous Malformation. We found 13 studies published from 1962 to 2015 (Table 1). A majority of cases were treated by surgery, and all articles reported a stable or improved clinical condition when the patient underwent treatment or conservative management.

Our case is a very particular case due to its clinical presentation. On the one hand, most AVMs associated with PTA presented as hemorrhagic stroke, either by intracerebral hemorrhage or subarachnoid hemorrhage. A small subset also presented as trigeminal neuralgia due to the location of the AVM [4, 8]. On the other hand, our case presented with epileptic seizures; to our knowledge, this is the only case of AVM with PTA that had this unusual clinical presentation.

The location of the AVMs is another point of discussion. Although they were once considered rare, the last published case reports have presented more posterior circulation AVMs, especially in the cerebellum and brainstem. Two of the reported AVMs were located in the corpus callosum; one had bilateral blood supply, and the other was only fed by the left circulation [9, 14]. Other sites where the AVMs were located include the occipital, temporal, and parietal regions.

Almost all the described associations of AVMs with PTA were incidental. In two cases, the PTA provided a direct blood supply to the AVM. In one case, the PTA was the main pedicle to an AVM located on the left side of the cerebellum [8], whereas in a second case, the PTA fed a right cerebellopontine AVM along with the ipsilateral anterior inferior cerebellar artery (AICA) [4].

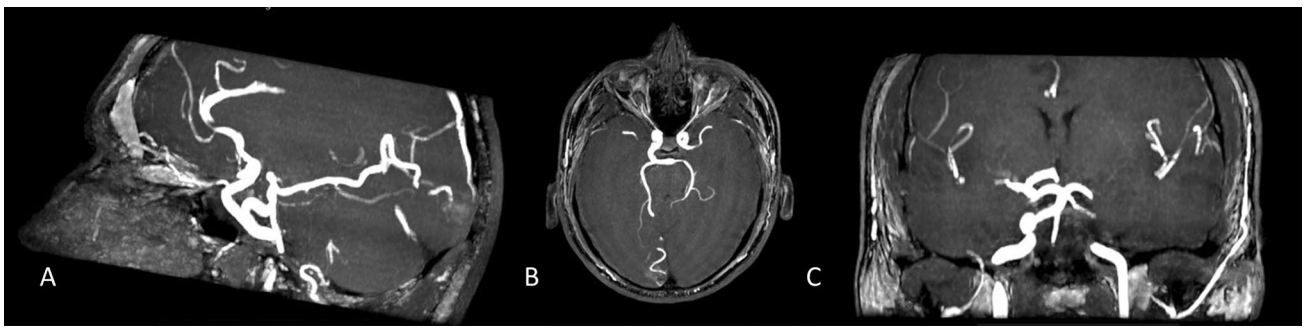

Fig. 1 Three-dimensional time-of-flight magnetic resonance angiography of the brain in sagittal (**A**), axial (**B**) and coronal (**C**) reformatted images show the presence of the persistent trigeminal artery arising from the right internal carotid artery and ending in the vertebrobasilar circulation. In the sagittal image, the right calcarine artery as well as the arteriovenous nidus can be identified

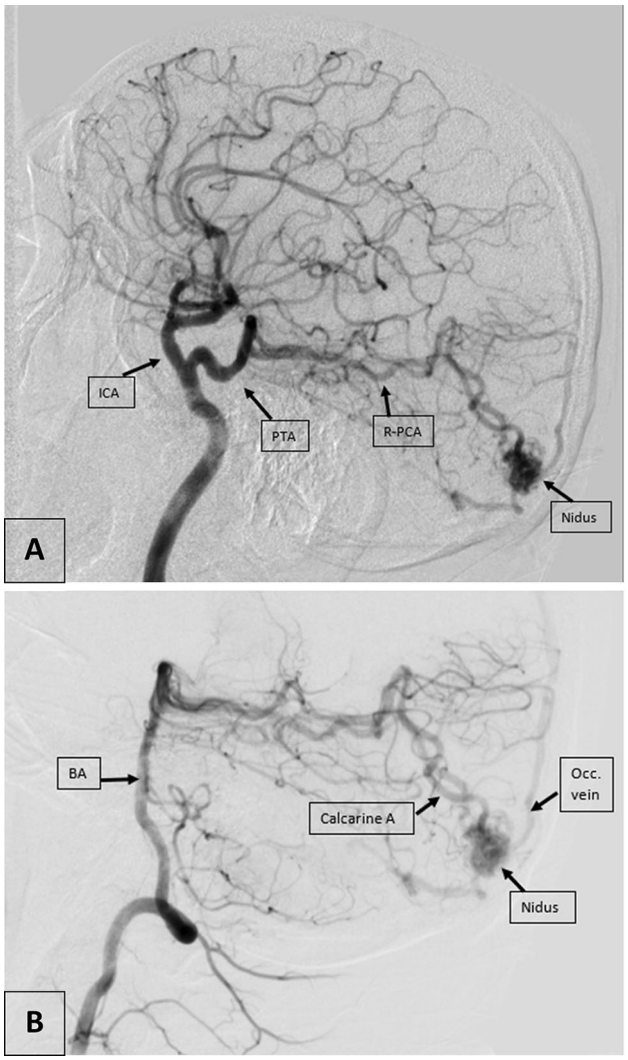

Fig. 2 Lateral projections of digital subtraction angiography of the right internal carotid (**A**) and vertebral (**B**) arteries show the presence of the persistent trigeminal artery (PTA) arising from the cavernous segments of the right internal carotid artery (ICA) and ending in the posterior circulation, from which the right posterior cerebral artery (R-PCA) arises. Vertebral angiography shows the basilar artery (BA) bifurcating into the posterior cerebral arteries; the right PCA divides into the right calcarine artery (Calcarine A) which feeds the AVM nidus (Nidus). The nidus drains into the Right Occipital Vein (Occ. vein)

Although a strong association of PTA with saccular aneurysms has been described, no formal studies have analyzed this phenomenon with AVMs. It has been theorized that the abnormal blood flow caused by AVMs may create an environment in which the PTA cannot be closed at birth [10]. Another theory states that both the failure to close the trigeminal artery and the emergence of the AVM are the result of maldevelopment in the fetal period [1]. This may be supported by the fact that most of the reported AVMs were located ipsilateral to the anatomical variant. Despite this, some dismiss these findings as purely coincidental.

Treatment of these malformations remains a difficult challenge. AVMs can be treated by surgical resection, endovascular embolization, and radiation surgery. In the case of endovascular treatment, PTA presents both an advantage and a disadvantage.

PTA acts as a communication mechanism between the anterior and posterior circulation. This elevates the risk of vessel occlusion when presented with embolizing material backflow [8]. However, in this particular case, PTA may also serve as an access pathway by which AVM catheterization may occur. Therefore, when anatomical variants are encountered, a thorough characterization of the anatomy using a diagnostic angiogram must be done before discussing treatment options.

Table 1 Summary of cases of cerebral arteriovenous malformations (AVMs) associated with persistent trigeminal artery (PTA)

References	Age, sex, country	AVM location	Trigger symptoms	Clinical diagnosis	Diagnostic study	Treatment	Outcome	Estimated final mRS[a]
Gannon [5]	46, female, USA	Right parieto-occipital	Headache and weakness	ICH	Angiogram	Surgical removal of AVM and hematoma	Clinical improvement. Persistent hemiparesis	3
Perret and Nishioka [12]	Not stated	Right pons	Not stated	ICH	Angiogram	Surgical entrapment of PTA	Clinical improvement. Symptom free at 36 months	0
Jayaraman et al. [6]	27, female, USA	Left temporal	Headache, vomiting, neck stiffness	ICH	Angiogram	Conservative	Normal neurologic function at 36 months	0
Tomsick et al. [13]	Not stated	Right temporal	Not stated	SAH	Angiogram	Uncertain	Uncertain	N/A
Brick and Roberts [3]	41, woman, USA	Right occipital	Headache, nausea, vomiting, back pain	ICH	DSA	Surgical resection	No recorded adverse events	0
Uchino et al. [14]	16, female, Japan	Corpus callosum, left arterial supply	Headache and vomiting	SAH	CT, DSA	Radiation therapy	Asymptomatic	0
Abe et al. [1]	48, female, Japan	Right parietal	Loss of consciousness, headache, vomiting	SAH	CT, DSA	Surgical resection	Recovered from neurological deficit	0
Nakai et al. [10]	53, male, Japan	Left cerebellum	Headache and vomiting	SAH	CT, DSA	Conservative	Clinical improvement after 3 months	0
Oran et al. [11]	29, male, Turkey	Left occipital	Headache, vomiting, neck stiffness	SAH, ICH	CT, DSA	Patient refused treatment	No follow up	
Mohanty et al. [9]	13, male, India	Corpus callosum, bilateral arterial supply	Headache, vomiting, loss of consciousness	Corpus callosum bleeding/intraventricular hemorrhage	CT, MRI, DSA	Gamma-Knife radiosurgery	Asymptomatic for 14 months	0
Kono et al. [8]	53, male, Japan	Left cerebellum fed by the PTA	Already diagnosed	Trigeminal neuralgia	CTA, MRI, DSA	Endovascular coiling of the distal PTA	Pain free for 12 months	0
Choudhri et al. [4]	64, male, USA	Right cerebellopontine	Sharp facial pain	Trigeminal neuralgia	MRI, post-surgical DSA	Surgical decompression + Cyber-Knife radiosurgery	Asymptomatic	0
Present case	21, male, Mexico	Right occipital	Seizures	Structural epilepsy	CTA, DSA	Medical treatment for epileptic seizures. Watchful waiting	Seizure free for 6 months	1

[a]The mRS was calculated based on the clinical descriptions provided in the articles and case series after treatment, either partial or definitive was undergone

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Availability of data and materials Clinical data related to the case are available upon request**.**

Declarations

Conflict of interest The authors declare no competing interests.

Ethics approval All procedures were carried out in accordance with the ethical standards of the Institutional Research Board, The General Health Law in Mexico and the Helsinki Declaration. The study was approved by the Internal Research Board.

Consent to participate Informed consent was obtained from the patient.

Consent for publication The patient consented to the use of their personal data included in this case report.

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