Persistent trigeminal artery in a patient with posterior circulation stroke treated with rt-PA: case report

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Abstract

Background: A persistent trigeminal artery (PTA) is a non-involuting embryonic vessel that connects the cavernous part of the internal carotid artery with the posterior circulation. In the adult it is associated with multiple pathological conditions including trigeminal neuralgia, ophthalmoplegia, hypopituitarism, intracavernous fistula, brain aneurysms and posterior circulation strokes. The latter may occur through steal phenomena or thrombosis in the anterior circulation. PTA associated vertebrobasilar hypoplasia has yet to be associated to TIA like events, however, in the reported case, that seems to be the case with reported vertigo being probably linked to vertebrobasilar insufficiency.

Case report: We present a case of an 82-year-old man with sudden onset neurological deficits, including left hemiparesis with crural predominance, vertical nystagmus, right internuclear ophthalmoplegia, dysarthria and dysmetria on the left arm. CT angiography disclosed basilar artery hypoplasia in the proximal two thirds and a persistent trigeminal artery. He was diagnosed with acute ischemic stroke. He was submitted to rt-PA with partial reversion of deficits.

Conclusion: The ischemic events related to PTA remain a rare cause of stroke with specific pathophysiological mechanisms and implications. They may occur through steal phenomena or thrombosis in the anterior circulation. Upon literature review, in the described case both mechanisms seem possible, however the transient episodes of vertigo could have been the first sign of vertebrobasilar insufficiency.

Keywords: Posterior trigeminal artery, Stroke, Vertebrobasilar insufficiency, Saltzman classification, CT angiography

Background

The trigeminal arteries have their origin in the embryonic vessels that connect the cavernous portion of the developing internal carotid arteries (ICA) and the paired longitudinal neural arteries that will later form the basilar artery (BA) [1, 2]. While the trigeminal artery usually involutes after the development of the posterior communicating artery (PcoA) there are cases, for reasons still unclear, where it remains persistent [2]. The PTA is the most common persistent embryonic carotid–basilar anastomosis [1]. Prevalence varies from 0.12 to 1% in studies using magnetic resonance angiography imaging or classical angiography [3–8]. In this paper we will briefly explain what is a PTA and to what pathologies it can be associated, we will review the meaning of a PTA in a posterior circulation stroke through a clinical case and we will present a table that summarizes the PTA related strokes reported in literature.

The different variations of the PTA can be cataloged using the Saltzman classification (Fig. 1), [1, 9, 10]. In Saltzman type 1, also called fetal PTA, the PTA insertion in the BA is distal to the anterior inferior cerebellar artery (AICA) and proximal to the superior cerebellar artery (SCA) and, in some cases, the BA proximal to the insertion of the PTA may be hypoplastic and the PcoA of the same side may be absent. In the Saltzman type 2 there is usually no hypoplasia of the BA, the PTA inserts proximally to the SCA, supplying them, and the PCAs are predominantly supplied by the PcoA. In the...
Saltzman type 3 variant, the PTA inserts directly into one of the cerebellar arteries, without having an anastomosis with the BA. In the case of Saltzman type 3 there are 3 variants: the type 3a variant that terminates in SCA; the type 3b variant, and the the most common one, that terminates into AICA; and type 3c variant that terminates into posterior inferior cerebellar artery (PICA) [1, 2]. In one study of 4,650 patients that underwent brain MRA, the prevalence of each type using the Saltzman classification was as follows: type I, 24%; type II, 16%; type III, 60% [7].

The PTA is linked to several pathologies, including vascular nerve compression syndromes, like trigeminal neuralgia (more prevalent in patients with PTA) [11, 12] or ophthalmoplegia due to oculomotor or abducens palsy [12–14]. It is also linked to hypopituitarism due to compression of the pituitary stalk [14, 15], spontaneous or traumatic intracavernous fistula [16] and brain aneurysms. Although there is no agreement that brain aneurysms are more prevalent in patients with PTA [4, 7, 17], the PTA itself is prone to aneurysms due to its bifurcation [1]. Finally, the PTA is also related to ischemic stroke, although, to our knowledge it has never been linked to TIA by vertebrobasilar insufficiency [18–30]. We present in Table 1 a structured summary of the PTA related stroke cases reported in literature.

**Case presentation**

An 82-year-old man, presented to the emergency department, on February 2018, after sudden onset of left hemiparesis and vertigo. Symptoms started during a period of greater physical effort, upon participation in a zumba class. His past medical history included transient episodes of vertigo during exercise in the previous months, an anterior circulation right hemispheric stroke in 2015, that left no sequelae, and hypertension controlled with a combination of 10 mg lisinopril and 2.5 mg amlodipine. The neurologic examination revealed mild left hemiparesis with facial involvement and crural predominance, vertical nystagmus, right internuclear ophthalmoplegia, dysarthria and dysmetria on the left arm. The total National Institutes of Health Stroke Scale (NIHSS) score was 6.

The plain brain CT scan was normal and a CT angiography showed hypoplasia of both vertebral arteries, the left terminating as the PICA, while the right gave origin to the BA. The BA had a filiform aspect in its proximal two thirds, having a normal caliber in the distal remaining third, after receiving a communicating artery from the cavernous segment of the left internal carotid artery, a PTA (Fig. 2). No abrupt stop of flow was identified. Echocardiogram showed severe dilation of the left auricle, as well as mild dilation of the right auricle and a 35 mm dilation of the proximal portion of the ascending aorta. Electrocardiogram was normal and a 48 h cardiac telmetry monitoring didn’t show any periods of arrhythmia. Lipid profile showed borderline high low density lipoprotein level, at 139 mg/dL.

The patient underwent treatment with recombinant tissue plasminogen activator (rt-PA) at two hours of symptom onset, this decision was based on the clinical presentation and in the absence of hemorrhage in the CT scan. Shortly after treatment the NIHSS score was 2, maintaining a slight paresis of the left leg and dysmetria on the left arm. Months later, the patient showed no neurologic sequelae.

**Discussion and conclusions**

PTA can be associated with many different vascular events. Patients with PTA and vertebrobasilar hypoplasia have a tendency to have a decreased vascular supply to the posterior fossa [19], this fact renders them susceptible to ischemic events. In this context, in the case of a stenosed carotid artery, a steal phenomenon can occur, and this can lead either to vertebrobasilar insufficiency or hemodynamic brain
| Identification | Symptoms                                                                 | Territory Confirmed by neuroimaging                                                                 | Arteries involved and saltzman classification of PTA | Treatment       | Patient outcome               |
|----------------|--------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------|---------------------------------------------------|-----------------|------------------------------|
| 42yo F (2010)  | Headaches, visual disturbances, right-sided numbness progressing to dysarthria and right-sided motor deficit with central facial palsy | Left anterolateral pontine infarction                                                              | Internal carotid artery (ICA) and PTA occlusion Saltzman type 1 | Heparine        | Residual right motor deficit |
| 47yo M (1992)  | Right hemiparesis and numbness of the left face                          | Ventral pontine lacunar infarct                                                                       | Hypoplasia of the VAs and VB Saltzman type 2       | Urokinase       | Hemiparetic gate             |
| 54yo M (2006)  | Aphasia, dysarthria and right hemiparesis                                | Left middle cerebral artery and posterior cerebral artery territories                                  | Embolization to the posterior circulation through the PTA Saltzman type 1 | None            | No deficits                  |
| 55yo M (2010)  | Dysarthria and right hemiparesis (grade 4/5)                             | Left ventral hemipontine infarction                                                                   | Ipsilateral tortuous primitive trigeminal artery Saltzman type 1 | Unknown         | Unknown                      |
| 56yo M (2014)  | Transient diplopia and right-sided numbness which resolved in 10–15 min, 2 days later horizontal nystagmus and diplopia, right hemisensory loss to light touch and pinprick, tongue deviation to the right, decreased fine motor function in the right hand and slow right finger-to-nose performance | Left ventral pons and left superior cerebellar peduncle                                      | Thrombosis of a persistent left trigeminal artery Saltzman type 1 | None            | Unknown                      |
| 58yo M (2016)  | Retrograde and anterograde amnesia, superior homonymous quadantanopias, and could not identify colors | Bilateral occipital infarcts involving the parahippocampal and lingual gyri                          | Hypoplastic vertebralbasilar circulation, with a persistent right trigeminal artery supplying the rostral basilar artery Saltzman type 1 | Unknown         | Unknown                      |
| 62yo F (2006)  | Transient ill-defined visual disturbance and a mild right-sided hemiparesis | Left mesencephalic                                                                                   | Right high-grade ICA stenosis (80%) hypoplastic vertebral arteries, right PTA Saltzman type 1         | ICA endarterectomy | No deficits                  |
| Identification | Symptoms | Territory Confirmed by neuroimaging | Arteries involved and saltzman classification of PTA | Treatment | Patient outcome |
|---------------|----------|--------------------------------------|---------------------------------------------------|-----------|----------------|
| 63yo M (1998) [23] | 1. Left facial palsy, right hemiparesis and left medial longitudinal fasciculus syndrome. 2. Hemiparesis and hemihypesthesia including pain and touch on the left side. | 1. Left basal part of thepons 2. Right basal part of the pons | Basilar artery middle portion stenosis (80%) Saltzman type 1 | 1. None 2. Urokinase | 1. Slight left facial palsy and right hemiparesis 2. No new deficits |
| 67yo M (2015) [26] | Sudden loss of consciousness and quadriplegia | Right cerebral hemisphere | Right middle cerebral artery (MCA) and BA occlusion Saltzman type 1 | Thrombectomy of the MCA and of the BA through the PTA | Lucid and with moderate left hemiparesis |
| 67yo M (2015) [27] | Sudden loss of consciousness and left hemiparesis | None | Right ICA occlusion with PTA, BA, posterior cerebral artery and superior cerebellar artery hypoperfusion Saltzman type 1 | rt-PA and thrombectomy through the ICA and PTA | Mild facial and left hand paresis |
| 70yo M (2010) [28] | Dysarthria and right hemiparesis | Left pontine infarction | Embolic occlusion of the PTA Saltzman type 1 | rt-PA | No deficits |
| 73yo M (2014) [29] | Weakness of the lower extremities, and blindness of the left eye | Multiple infarcts in the anterior and posterior circulation (cardioembolisms) | Persistent primitive trigeminal artery, basilar artery hypoplasia Saltzman type 1 | Unknown | Unknown |
| 76yo M (2016) [30] | Mild dysmetria and intentional tremor affecting the right arm, tandem gait ataxia, and right-sided hemianopia, preceded several weeks by intermittent vertigo | Border-zone territory between the MCA and the posterior cerebral artery (PCA) | Left ICA stenosis (75%) and left PTA, BA hypoplasia below the PTA Saltzman type 1 | ICA endarterectomy | Resolution of the intermittent vertigo |
| 80yo F (2010) [29] | Dysphasia and dysarthria with dizziness and cough | Right pontine infarct | PTA between left vertebral artery and ipsilateral external carotid artery, right vertebral artery hypoplasia Saltzman type 2 | Unknown | Unknown |
stem infarction [31]. In the case of BA occlusion however, if the occlusion is proximal to the insertion of the PTA, the PTA can have a protective effect in the distal territory [25]. In the case of an anterior circulation thrombus, the PTA can lead to migration of the thrombus to the BA, possibly causing a posterior circulation stroke [20, 24]. Finally, in BA hypoplasia, PTA also opens the possibility for thrombectomy in the posterior circulation [26, 27].

Despite all the variety of ischemic events related with PTA, thrombosis of this vessel remains a rare phenomenon, although there is a case report of PTA thrombosis related with internal carotid dissection [18].

Our patient had a congenital Saltzman type 1 PTA variant and presented with symptoms suggestive of acute brainstem infarction that partially resolved after the administration of rt-PA. The clinical manifestations are compatible with an ischemic lesion of the basis pons, causing an ataxic hemiparesis syndrome, and extending to the tegmental region, affecting the longitudinal medial fasciculus. This suggests that the most affected territory was dependent from the small perforating vessels of the BA. Furthermore, the patient was exposed to extenuating exercise when the symptoms started, this favors the possibility that the event was due to a steal phenomenon, but the fact that the patient had no great artery disease and the fact that the patient improved after rt-PA suggests that the mechanism could also have been thrombo-embolic. Upon literature review both mechanisms seem possible. We would also like to emphasize that the reported episodes of vertigo probably corresponded to previous transient ischemic attacks (TIA) in the same area, possibly being the first sign of vertebrobasilar insufficiency. To our knowledge this is the first case study to point out the possibility of having several transient episodes (in this case of vertigo) compatible with vertebrobasilar insufficiency as a premonitory sign of PTA related stroke.

**Abbreviations**

- AICA: Anterior inferior cerebellar artery
- BA: Basilar artery
- ICA: Internal carotid artery
- NIHSS: National Institutes of Health Stroke Scale
- PcoA: Posterior communicating artery
- PTA: Persistent trigeminal artery
- rt-PA: Recombinant tissue plasminogen activator
- SCA: Superior cerebellar artery
- TIA: Transient ischemic attacks
- VA: Vertebral artery

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**Competing interests**

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