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CASE REPORT

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Combination of Persistent Trigeminal Artery and Ischemic Stroke: Report of 2 Cases

Persistan Trigeminal Arter ve İskemik İnme Birlikteliği Olan 2 Olgu Sunumu

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ABSTRACT

Persistent trigeminal artery (PTA) is the most common embryological anastomosis between the internal carotid artery (ICA) and basilar artery, which continues also in adult life. PTA is mostly asymptomatic but has also been described with comorbid conditions such as cerebrovascular pathologies. In this article, two cases of ischemic stroke accompanied by incidentally detected PTA are presented. In the first case, ischemic stroke with ipsilateral ICA hypoplasia and PTA were observed and in the second case, ischemic stroke with ipsilateral critical ICA stenosis and PTA were observed. The presence of minor infarction and low disability in both cases suggests that the presence of PTA may be associated with a better prognosis in ischemic stroke as it supports collateral circulation.

Keywords: Basilar-carotid artery anastomoses, persistent trigeminal artery, ischemic stroke.

ÖZ

Persistan trigeminal arter (PTA), internal karotis arter (ICA) ve baziller arter arasında erişkin hayatta da devam eden en sık embriyolojik anastomozdur. Çoğunlukla asemptomatik olarak bilinen PTA, serebrovasküler patolojiler gibi eşlik eden durumlar ile de tanımlanmıştır. Bu makalede insidental saptanan PTA ve iskemik inme birlikteliğini konu alan iki olgu sunuldu. İlk olguda iskemik inme ile ipsilateral (IKA) hipoplazisi ve PTA, ikinci olguda ise ipsilateral kritik ICA darlığı ve PTA izlendi. Her iki olguda minör enfarkt ile prezentasyon ve dizabilitenin az olması PTA varlığının kollateral dolaşımı desteklemesi sonucu iskemik inmede iyi prognoz ile ilişkili olabileceğini düşündürmektedir.

Anahtar Kelimeler: Baziler-karotid arter anastomozları, persistan trigeminal arter, iskemik inme.

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CASE PRESENTATION

Carotid-vertebrobasilar anastomoses, which supply blood flow from the internal carotid artery (ICA) to the vertebrobasilar system during embryonic development, are called presegmental arteries.[1] The presegmental arteries disappear with the involution in the intrauterine period, but if there is a problem in this process, they do not regress and can be seen in adulthood and are named with the names of the cranial nerves that accompany them.[1,2] The most common presegmental arteries in adults are the persistent trigeminal artery (PTA), the persistent hypoglossal artery, the persistent otic artery and the persistent proatlantal intersegmental artery [1] (Figure 1). PTA is the most common of these, accounting for about 85% of all persistent presegmental arteries, and its prevalence has been reported to be 0.1%-0.6%.[1,3] The PTA, typically unilateral, connects the anterior and posterior circulations by diverting blood from the cavernous ICA to the longitudinal neural arteries, the precursors of the vertebral-basilar system.[3,4] Almost all cases are incidental radiological findings and usually asymptomatic.

However, it has also been described in the literature with cerebrovascular pathologies such as aneurysm, trigeminocavernous fistula and thromboembolic ischaemia and associated conditions such as cranial nerve palsy, trigeminal neuralgia, vertigo, ataxia, migraine and headache.[4,5] In this article, two cases of incidental PTA and ischaemic stroke are presented.

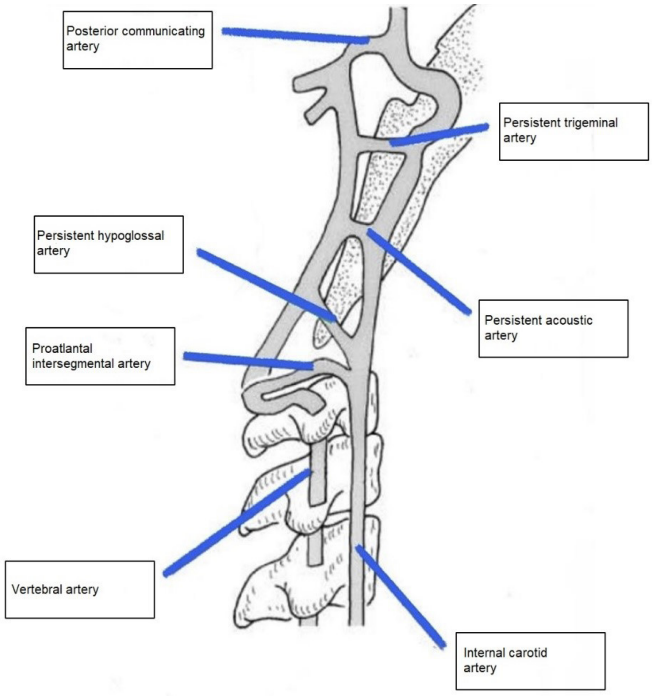

Figure 1. The most common presegmental arteries in adulthood.

Case 1

A 57-year-old left-handed female patient was admitted to the emergency department with complaints of slurred speech, difficulty understanding what was being said and difficulty walking since the morning. She smoked 36 packs/year and had no known chronic diseases except for migraine diagnosed 20 years ago. It was found that her previous migraine attacks presented with throbbing headache, nausea and vomiting in the right side of the head and the last attack with a similar character was experienced in the morning during the event. Neurological examination revealed mild paretic left upper and lower extremities and weak left basal reflex. Diffusion magnetic resonance imaging showed multiple T2 hyperintense lesions with acute diffusion restriction in the right insula and right frontoparietotemporal regions. The largest lesion was in the right insula (Figure 2).

MAIN POINTS

- PTA is the most common permanent carotid-vertebrobasilar anastomosis.
- Although PTA has been linked to cerebrovascular diseases, its role as a risk factor for stroke remains unclear
- PTA may be associated with a better prognosis in ischemic stroke, as it can support collateral circulation in cases of severe carotid artery stenosis or hypoplasia

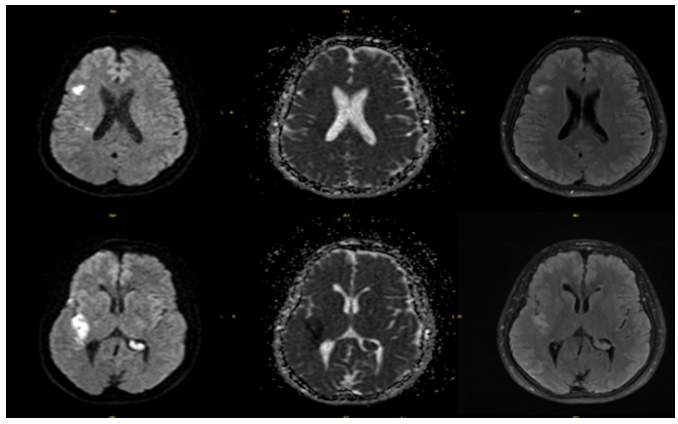

Figure 2. Acute diffusion restriction in the right frontoparietotemporal region and insula observed in DWI, ADC and T2 FLAIR sequences from left to right on diffusion MRI. Lesions are observed as hyperintense on T2 FLAIR imaging.

The patient who was admitted at the 12th hour of the incident and whose National Institutes of Health Stroke Scale Score (NIHSS) was calculated as 3, was referred to the neurological service. There was no cardioembolic focus in the examinations ordered for the etiology of stroke. Carotid vertebral doppler ultrasonography (CVDUS) revealed a severe stenosis extending from the right ICA bulb to the proximal ICA, suggesting atherosclerosis in the etiology of stroke. Carotid computed tomography angiography (CTA) revealed that in addition to the critical stenosis in the right ICA, both ICAs were thin calibre after proximal segment, more prominent on the right, posterior vascular structures were normal, but there was persistent trigeminal artery (PTA) variation between anterior and posterior system. Digital subtraction angiography (DSA) was planned and the direction of flow of the PTA was from the distal basilar artery to the ICA (Figure 3). Endovascular treatment was not an option and discharge was planned after dual antiplatelet and statin therapy. Verbal and written informed consent was obtained from the patient to share data.

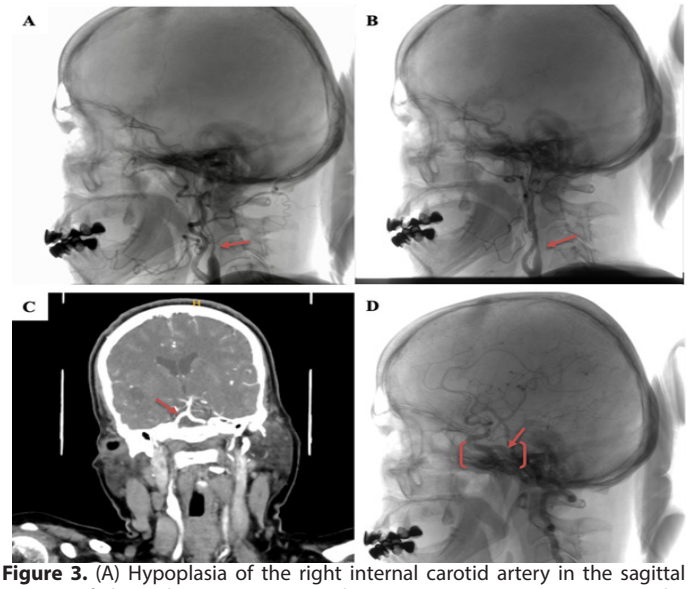

Figure 3. (A) Hypoplasia of the right internal carotid artery in the sagittal section of the right common carotid artery (CCA) injection. (B) Hypoplasia of the left internal carotid artery in the sagittal section of the left CCA injection. (C) Persistent trigeminal artery on the right in the coronal section of CT angiography. (D) Tau sign (red brackets) consisting of the anterior horizontal and vertical parts of the right ICA and the proximal part of the PTA in the sagittal section of the right CCA injection. Persistent trigeminal artery on the right (red arrow).

Case 2

A 64-year-old woman with a history of paroxysmal atrial fibrillation, hypertension, diabetes mellitus, chronic kidney disease, and an ischemic stroke 2.5 years ago presented with weakness in the left side of her body. She was examined at the stroke outpatient clinic. The neurological examination was normal. Current brain MR imaging of the patient showed multiple lacunar infarct areas prominent in the right hemisphere, and MR imaging during the ischemic stroke period showed millimetric diffusion restriction in the right cerebral hemisphere in the frontoparietotemporal, occipital, right centrum semioval, and right posterior external capsule (Figure 4). The follow-up CVUSG performed 2 years later found that the atherosclerotic changes that previously caused 30-35% stenosis at the right ICA entrance had progressed to cause more than 70% stenosis. Atherosclerotic changes causing a 90% stenosis at the narrowest part of the proximal part of the right ICA and a persistent variation of the trigeminal artery extending between the basilar artery and the apex of the right ICA were observed (Figure 5). An endovascular stent was placed in the right carotid artery and the patient was discharged with acetylsalicylic acid 100 mg 1x1 added to rivaroxoban 20 mg 1x1 treatment. Verbal and written informed consent was obtained from the patient to share data.

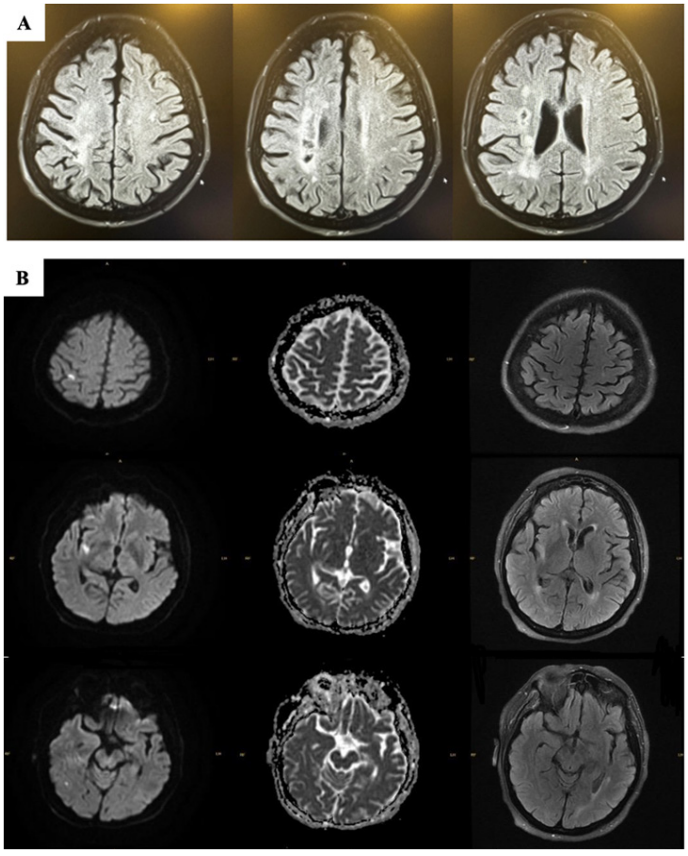

Figure 4. (A) Lacunar infarct areas in the right centrum semiovale on diffusion MRT2 FLAIR images. (B) Millimetric diffusion restrictions, some of which cannot be distinguished with ADC equivalents, in the right centrum semiovale, right external capsule and right occipital region, observed in DWI, ADC and T2 FLAIR sequences from left to right on previous diffusion MR imaging.

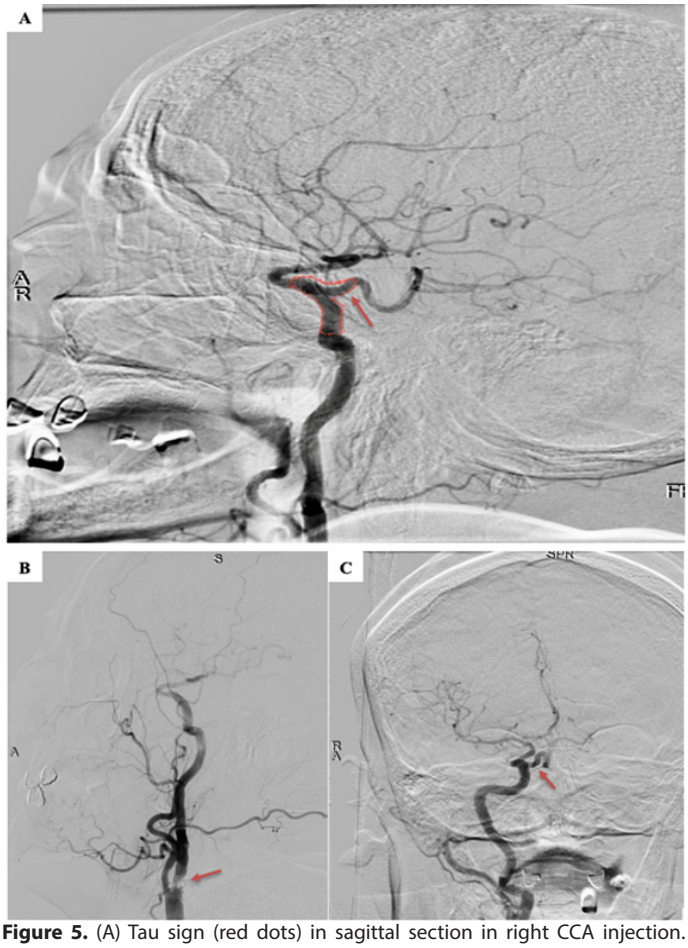

Figure 5. (A) Tau sign (red dots) in sagittal section in right CCA injection. Persistent trigeminal artery on the right (red arrow). (B) Stenosis in the right internal carotid artery in sagittal section in right CCA injection. (C) Persistent trigeminal artery on the right in coronal section in right CCA injection.

DISCUSSION

First reported by Quain in 1844 in an autopsy case and by Sutton in 1950 in an angiography case, PTA is the most common permanent carotid-vertebrobasilar anastomosis.[6] There are several classifications of PTA. These are based on the course of the artery, the type of termination, and the associated vascular pathology. Saltzman first identified two types of PTA in 1959. Saltzman type I, also known as fetal PTA, is characterized by PTA supplying the distal vertebrobasilar arteries. The posterior communicating artery, the basilar artery, and the distal vertebral artery may be absent or hypoplasic, respectively. Saltzman type II, on the other hand, is characterized by the PTA supplying the superior cerebellar arteries and the posterior cerebral arteries supplying the posterior communicating arteries. In Saltzman type II, the hypoplasia of the BA is usually not present. In the Saltzman type III variant, which for some time was considered to be a combination of type I and type II, the PTA enters directly into one of the cerebellar arteries without anastomosis to the BA. The classification was later expanded to five types by Weon et al.[9] Types 1 and 2 correspond to the classification of Saltzman, whereas in types 3 and 4, the PTA drains into the contralateral and ipsilateral posterior cerebral arteries, respectively. Type 5 can be subdivided into type 5a superior cerebellar artery (SCA), type 5b anterior inferior cerebellar artery (AICA), and type 5c posterior inferior cerebellar artery (PICA). Type 1 is reported to be the most common.[9] (Figure 6).

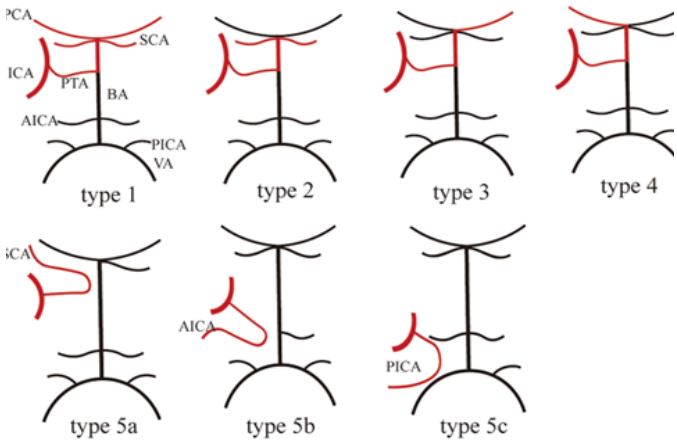

Figure 6. Classification of persistent trigeminal artery according to Weon et al.[9] ICA, internal carotid artery; SCA, superior cerebellar artery; AICA, anterior inferior cerebellar artery; PICA, posterior inferior cerebellar artery; BA, basilar artery; VA, vertebral artery; PCA, posterior cerebral artery.

In our cases, the PTA also showed direct drainage into the basilar artery and was classified as type 1, which is the most common type. The classic angiographic sign of a persistent trigeminal artery is the Tau (τ) sign. The sign, named for its visual resemblance to the letter Tau (τ) in the Greek alphabet, is the image formed by the anterior horizontal and vertical portions of the internal carotid artery and the posterior portion of the proximal trigeminal artery in sagittal images.[10] Tau sign was present in both of our cases (Figure 3 and 5).

The occurrence of ischemic stroke associated with PTA is rarely reported; among the few cases reported to date, most have been minor ischemic strokes, but the role of PTA in ischemic stroke is not well understood. Ferreira et al.[8] reported that patients with PTA and vertebrobasilar hypoplasia are more prone to ischemic stroke in the posterior system due to hypoperfusion in the posterior fossa.[1,8] However, there have been reports in the literature that the anterior and posterior systems were affected together or that the ischemic stroke was only in the anterior system.[11,12] In our cases, the first case had anterior circulation involvement and the second case had both anterior and posterior circulation involvement in the same hemisphere. The majority of case reports on the role of PTA in ischemic stroke support the view that the presence of PTA may be associated with an increase in the risk of ischemic stroke.[4] This can happen through two mechanisms. Weakened flow of the proximal basilar artery and bilateral vertebral arteries may play some role in posterior circulation stroke, or an anterior circulation thrombus may float freely along this persistent embryonic conduit into the posterior circulation, i.e., ulcerative atherosclerotic disease of the internal carotid artery may be indicated as a potential source of emboli reaching the posterior circulation via PTA.[4,13] In our second case, both anterior and posterior lesions were found, suggesting that the PTA-mediated embolus may have translocated to the posterior system. Physiologically, PTA is considered to flow from the ICA to the BA. However, flow may be reversed in congenital absence, severe stenosis, or occluded ICA.[5] Considering the presence of hypoplasia in both ICAs in our first case and the reversal of flow due to severe ICA stenosis in the second case, we can assume that the PTA is collateral. PTA may be beneficial in the presence of collateral circulation during stroke, such as leptomeningeal collaterals, external-internal carotid artery anastomoses, and the polygon of Willis, and infarct volume may decrease as the number of collaterals increases.[5] This is supported by the fact that our first patient had a minor stroke despite hypoplasia of the ICA and our second patient recovered without sequelae despite severe proximal carotid stenosis. It is also known that in certain cases of PTA with a large vessel occlusion, this alternative colleteral circulation can be used as a therapeutic pathway for mechanical thrombectomy.[14]

In 2019, Lyu et al.[15] first reported a patient with PTA and ipsilateral ICA distal anastomosis in whom MR angiography showed a posterior cortical border infarct. In 2022, Watanabe et al.[16] described a case of PTA and ipsilateral ICA hypertrophy presenting with chronic headache. Uhlig et al.[12] reported a case of PTA with migraine-like headache and left temporal lobe ischemic lesion. Our first case is probably the second reported case of acute ischemic stroke in association with PTA and ipsilateral ICA hypoplasia. Furthermore, the ipsilateral migraine attack presentation in this case is noteworthy because of the combination of clinical presentations that can accompany PTA. The second case of incidental detection of ipsilateral severe ICA stenosis by PTA is important considering the coexistence of multiple risk factors in patients with ischaemic stroke and the documentation of PTA and progressive ipsilateral ICA stenosis.

Finally, it is aimed to underline the association of rare PTA variation and ischemic stroke. Although there have been reported cases of stroke associated with PTA, it is still not fully known whether or not PTA is a risk factor for stroke. However, as observed in our cases, PTA can be detected incidentally in patients with infarcts in both anterior and posterior circulation, and it should be noted that PTA can contribute positively to the prognosis in ischemic stroke patients by increasing collateral circulation.

Informed Consent: Written informed consent was obtained from patients in this study.
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