A Rare Case of Persistent Primitive Trigeminal Artery with Multiple Anomalies of Cerebral Vessels

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To the Editor: A 38-year-old man was admitted to our hospital with transient aphasia and weakness of the right extremities. As a habitual smoker, he had been diagnosed with hypertension and hyperlipidemia 3 and 10 years prior, respectively. With regard to family history, he stated that his father had suffered cerebral infarction.

On admission, he was found to have abnormal blood pressure (150/90 mmHg; 1 mmHg = 0.133 kPa) and low-density lipoprotein level (3.8 mmol/L). Cranial magnetic resonance imaging (MRI) showed multiple ischemic lesions under both frontal cortices. Common carotid angiograms showed symmetrical anomalies in both external carotid arteries. The facial, lingual, and superior thyroid arteries arose from the ipsilateral common carotid artery [Figure 1a and 1b]. Right internal carotid angiogram showed complete occlusion of the right middle cerebral artery and the formation of moyamoya vessels in the basal ganglia and a persistent primitive trigeminal artery (PPTA) that branched from the C4 portion of the right internal carotid artery and terminated at the upper portion of the basilar artery [Figure 1c]. Left internal carotid angiogram revealed occlusion of the left middle cerebral artery and the formation of a few moyamoya vessels [Figure 1d]. Left vertebral artery angiogram showed that the posterior circulation system provided the compensatory blood supply to the anterior circulation through bilateral posterior communicating arteries and distal cortical branches [Figure 1e]. Computed tomography angiography also revealed the incidental finding of an associated fenestration of the basilar artery [Figure 1f]. The patient was treated conservatively with medications and was discharged two weeks later. His symptoms have not recurred during the follow-up period of one year.

PPTA is a temporary embryonic vascular anastomosis between the carotid and vertebrobasilar arteries, with a reported incidence of 0.03–2.20%.[1] There is a female sex predilection, and it may occur in patients of any age.[2] PPTA generally causes no specific symptoms and occasionally presents with other cerebrovascular variants.[3] Only 14 patients with anastomosis in association with moyamoya disease have been reported, with initial symptoms mainly caused by cerebrovascular diseases.[3,4] Our case was diagnosed as a transient ischemic attack based on clinical symptoms and cranial MRI, most likely caused by low cerebral blood perfusion due to moyamoya disease. The first and only case of the association of PPTA and fenestration of the basilar artery was reported in 1997 by Hattori *et al*.,[5] who described a 39-year-old man with acute putaminal hemorrhage with the coexistence of both anomalies. The PPTA in our patient was not only combined with moyamoya disease and fenestration of the basilar artery but also with consolidated symmetrical variations of branches of both external carotid arteries, which has never been reported. What are the clinical implications about these abnormal vascular structures?

Although we did not find their inner links among the cerebrovascular anomalies appeared simultaneously in the same patient, we considered this was not an isolated or occasional clinical phenomenon. These vascular anomalies may mainly be associated with abnormal embryonic development, suggesting that numerous variations are possible in cerebral artery development in the human embryo; they may further help us understand the specific cerebrovascular variations and anatomy. Future studies should focus on the existence of such diseases. This would be very useful in the development of *in vivo* models for researching the pathogenesis of these vascular anomalies.

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Conflicts of interest

There are no conflicts of interest.

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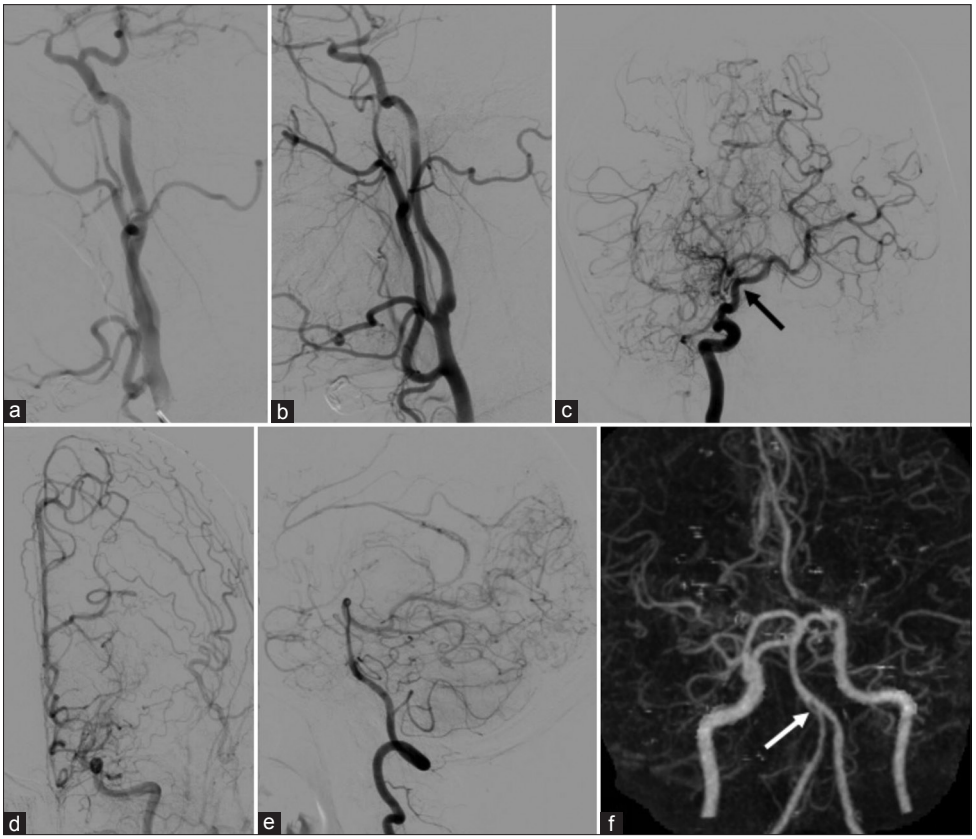

Figure 1: (a and b) Common carotid angiograms showed that bilateral facial artery, lingual artery, and superior thyroid artery originated from the ipsilateral common carotid artery; (c and d) internal carotid angiograms showed the complete occlusion of both middle cerebral arteries and the formation of moyamoya vessels and a persistent primitive trigeminal artery (black arrow); (e) left vertebral artery angiogram showed the collateral circulation established from the posterior circulation to the anterior one; (f) computed tomography angiography showed the fenestration of the basilar artery (white arrow).

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