Arteriovenous Malformation and Persistent Trigeminal Artery Association; Case Report

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Abstract: Persistent trigeminal artery that leads blood flow rostrocaudally between the primitive internal carotid artery and the parallel longitudinal neural plexus during embryogenic development, may sometimes be associated with other vascular anomalies due to the same congenital developmental cerebrovascular disturbance. Persistent trigeminal artery and aneurysm combination is most frequently seen, but the association with arteriovenous malformation is rare. A 12-year old girl with a right parietal arteriovenous malformation which led to intracerebral and intraventricular haemorrhage, was admitted with a left hemiparesis involving the face. After successful excision of the arteriovenous malformation, the left hemiparesis improved and persistent trigeminal artery had disappeared on late control carotid angiogram. This rare association is discussed with the relevant literature.

Key words: Carotid-basilar anastomosis, cerebral arteries, persistent trigeminal artery, vascular malformations.

INTRODUCTION

An anomalous anastomotic vessel between the internal carotid artery (ICA) and the basilar artery was first reported by Quain in an autopsy case in 1884. Later this vessel was named trigeminal artery by Padget (11). The first angiographic demonstration of persistent trigeminal artery (PTA) was made by Sutton in 1950 (14). In addition, three embryogenic carotico-basilar anastomoses, persistent hypoglossal artery, persistent otic artery and persistent proatlantal artery have been reported by other authors. Although arteriovenous malformations (AVM) are congenital lesions, they are not frequently seen with other vascular abnormalities except aneurysms. The rate of AVMs associated with other vascular abnormalities is 2.7-16.7% and the contribution of persistent embryogenic carotico-basilar anastomoses extremely rare. In this report, we present a case with such a rare combination of vascular anomalies and discuss the role of haemodynamics in the persistency of PTA (18).

CASE REPORT

A 12-year old girl was admitted with sudden left upper and lower extremity weakness in 1989. She complained of intermittent headache and vomiting for two years. There was no seizure history. Neurological examination showed a severe left hemiparesis. Because a right frontoparietal intracerebral haematoma which opened to the lateral ventricles was observed on computed tomographic scans, percutaneous carotid angiography was performed. A right parietal AVM supplied via a branch of the middle cerebral artery and a right PTA were visualized. There was well-calibrated patency, enough to transmit blood flow from the right ICA to the posterior circulation via the right posterior communicating artery (PCoA) and PTA. At operation two days later, after the evacuation of the haematoma, the nidus of the malformation was excised totally with standard microsurgical techniques. No complication or seizure developed in the postoperative period. The left hemiparesis improved
over three weeks. Control right carotid angiogram performed one month after the operation showed that AVM was totally excised and the PTA was not patent, but the right PCoA was still working well. There were minor focal electroencephalographic abnormalities in the operation region and carbamazepine was prescribed for one year. The latest neurological examination six years postoperatively, revealed no neurological deficit or learning difficulties.

**DISCUSSION**

During the stage of 3mm, while the forebrain of the embryo is fed by primitive ICA which are the cranial extensions of the paired dorsal aorta, the parallel longitudinal neural plexuses created by anastomosis of the cranial segmental arteries, appear on the ventrolateral aspect of the hindbrain (8). These plexuses later form the basilar artery (2). At this stage, rostrocaudal blood flow from the carotid arteries to the parallel longitudinal plexuses is secured by four significant anastomotic vessels. Among these, three are cranial segmental and one is a suboccipital-intersegmental. These carotid-basilar anastomotic vessels are named according to the homologous adjacent cranial nerves or vessels. PTA is the most important and is the main vessel that transmits blood to the hindbrain for a short period. At the stage of 5-6mm (29 days), the rostral part of each longitudinal neural plexus has permanent communication via a primitive posterior communicating artery from the posterior trunk of the primitive trigeminal artery. The caudal end of every longitudinal plexus reaches the cervical region and anastomoses with the primitive vertebral arteries ascending from longitudinal anastomotic vessels of cervical intersegmental
Fig 3: During the capillary phase of the right carotid angiogram, a right frontoparietal AVM was visible on lateral projection.

Fig 4: In the arterial phase of the postoperative right carotid angiogram, A) PTA could not be visualised on AIP view. B) Total excision of the AVM was proven on the lateral view of the capillary phase.

arteries which are branches of the dorsal aorta. At the stage of 11.5mm (34 days) basilar and vertebral arteries are completely formed (2) and carotidbasilary anastomotic vessels begin to be obliterated and disappear at the stage of 14mm (6). These anastomoses are patent for a period of seven to ten days (8). But in rare cases this spontaneous obliteration fails and caroticobasilar anastomotic vessels persist. Among the published cases of persistent caroticobasilar anastomotic vessels, PTA constitutes 85% (3). PTA usually originates as soon as the ICA emerges from the carotid canal, and passes near or directly through the cavernous sinus to the tip or distal one third of the basilar artery (1). In our case, the origin of the PTA was the same, but it terminated on the middle third of the basilar artery and was associated with a well working ipsilateral PCoA that led to visualisation of the posterior circulation via a left carotid injection. Failure in the obliteration of the PTA and the efficiency of the PCoA might be due to blood stealing to the AVM nidus.

The incidence of PTA in angiograms is 0.1-0.2% (8). The cerebrovascular disorder that leads to PTA may be associated with AVM, Moya Moya disease and in 25% of cases, with aneurysm. But the incidence of PTA in cases with AVM is very low. The first case was published by Krayenbühl and Yaşargil in 1957. Jayaraman et al., reviewing the literature, found only 11 cases up to 1977 and added their case as the twelfth (5). Then in 1987, Yaşargil reported that he had not seen any PTA in his AVM series of 500 cases (18). Uchino et al., reported another case of corpus callosum AVM associated with PTA in 1989 (15).

Some authors suggest that in states of vascular insufficiency due to embolus or stenosis, persistent
carotocobasiliary anastomotic vessels may lead to a haemodynamic change in favour of the ischaemic region and improve survival (1). But a well working anastomotic vessel may be associated with special clinical findings. PTA may be the atypical reason for facial neuralgia (4, 7, 10), hemifacial spasm (7) and oculomotor nerve dysfunction (9, 10, 17). Also, these vessels may be a route for microemboli which arise from the carotid plaques and reach the posterior fossa circulation (12, 13, 14). In our case, we observed no such clinical complaints or findings. The patient was admitted to hospital with severe left hemiparesis preceded by a sudden headache due to bleeding of the AVM. The patency of the PTA and the well-working PCoA may contribute to blood being taken from the posterior to the anterior circulation and reaching critical pressure and flow level through the nidus before bleeding. But, maybe this communication also led to an increase in the blood supply of the damaged parenchyma since neurological improvement started in the early postoperative period. Disappearance of the PTA on the late angiogram may be explained by the lack of blood stealing by the anterior circulation after total excision of the AVM.

We believe that the appearance of a persistent caroticobasiliary anastomosis in cases with aneurysm or AVM, is not an insignificant incidental finding. It not only explains the haemodynamics of the lesions, but also precludes alternative treatment modalities. Detailed evaluation should include the role of persistent embryogenic vessels in the haemodynamic characteristics of vascular disorders.

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