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Case Report

Persistent Trigeminal Artery Associated with Double Aneurysm

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Abstract

In this manuscript, we present a rare case of the persistent trigeminal artery (PTA). PTA is the most common of the primitive embryonic anastomosis, the incidence is very rare, approximately 0,2% and may associate with aneurysms and vascular anomalies. A primitive embryonic anastomosis (i.e. hypoglossal artery, proatlantal intersegmental artery, otic artery, and trigeminal artery) could persist into adulthood. Here, we report a 53 years old female with right side ptosis and diplopia associated with a double aneurysm, PTA and bilateral vertebral artery hypoplasia which was found during the angiography and successfully treated with endovascular coiling. The finding in these cases may shed light on clinical presentation and give an aid for the treatment of this clinical setting.

Keywords: *Persistent Trigeminal Artery; Aneurysm; Internal Carotid Artery; Posterior Communicans Artery; Angiography; Coiling*

Abbreviations

PTA: Persistent Trigeminal Artery; CA: Carotid Artery; ICA: Internal Carotid Artery; BA: Basilar Artery; SCA: Superior Cerebellar Artery; PCA: Posterior Cerebral Artery; AICA: Anterior Inferior Cerebellar Artery; PCom: Posterior Communicans; MRI: Magnetic Resonance Imaging; MRA: Magnetic Resonance Angiography; DSA: Digital Subtraction Angiography

Introduction

In 1844, a persistent trigeminal artery (PTA) was firstly described by Quain after he performed an autopsy. And in 1950, the first angiography technique was reported by Sutton [1-3]. PTA is a rare anastomosis between carotid artery (CA) and basilar artery (BA) and has incidence between 0.1% to 0.3% [3,4]. The objective of this case report is to present a rare type of PTA based on brain magnetic resonance angiography (MRA) and digital subtraction angiography (DSA) and further management by endovascular coiling.

Case Report

We report a 53 years old female with right side ptosis, diplopia, and throbbing headache for a week. There was no history of head trauma, diabetes, and hypertension. The finding on physical examination at the time of admission to our hospital were as follows; body temperature 36.5oC; pulse rate 72 beats/min, respiratory rate 18 cycles/min and blood pressure recorded of 130/80 mmHg. She was conscious, Glasgow Coma Scale E4M6V5, and normal cognitive function. Examination of eye revealed right oculomotor and trochlear nerve palsies and there was no visual defect. Laboratory test including blood counts, biochemistry, lipid panel, and urinalysis was normal.

Based on magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) images, the brain revealed bilateral subcortical lacunar lesions and suspicion of thrombosis. The patient elected to undergo head digital subtraction angiography (DSA). DSA images showed unruptured saccular aneurysm which grew on bifurcation of the right internal carotid artery (ICA) and posterior communicans (PCom) artery with size 4.43 mm x 5.52 mm, and neck 1.65 mm (Figure 1). We also found unruptured saccular aneurysm on the proximal left PCom artery with size 7.48 mm x 5.52 mm, and neck 4.78 mm. This PTA grows from right ICA into BA and located between superior cerebellar artery (SCA) and anterior inferior cerebellar artery (AICA). This artery got blood supply directly from ICA and supplied basilar system and PCom artery. On this imaging, we also found hypoplasia in both vertebral arteries. Based on Saltzman classification, this PTA was classified as Saltzman type II.

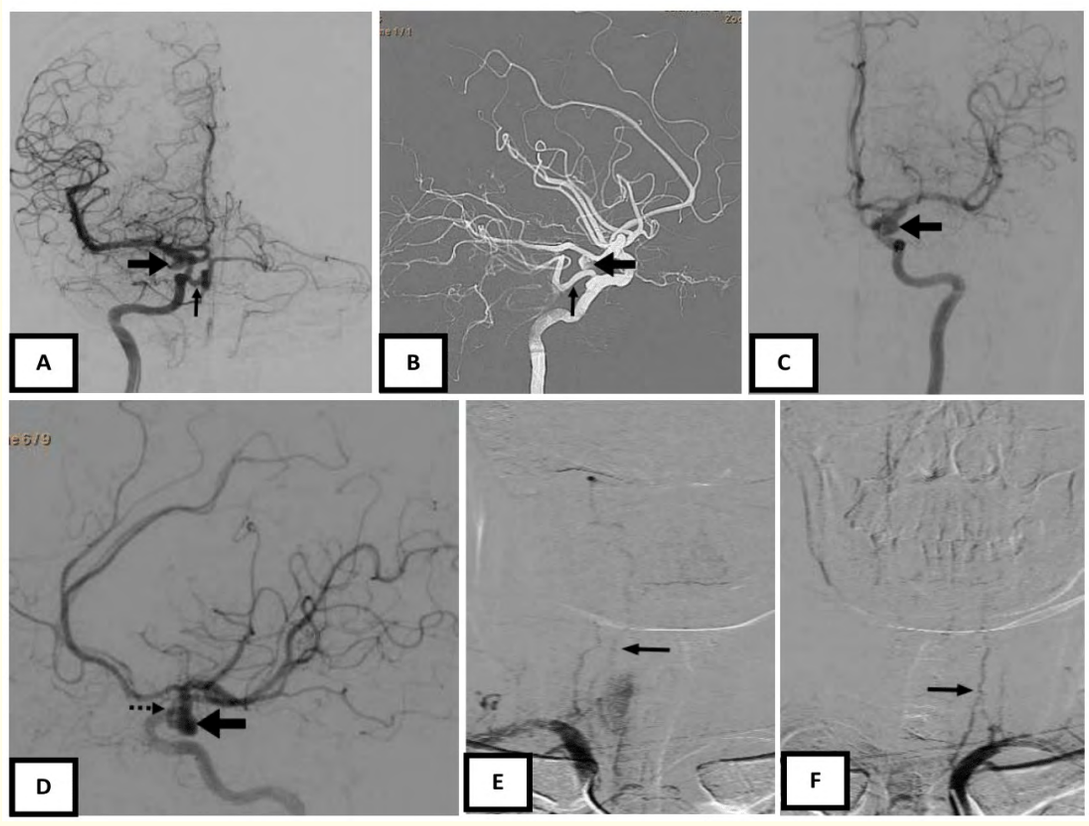

Figure 1: *Digital subtraction angiography (DSA) imaging on the patient. (A-B) The unruptured saccular aneurysm grew on the right bifurcation of right ICA and PCom artery (bold arrow). The PTA grows from an internal carotid artery (ICA) into a basilar artery (BA), and located between superior cerebellar anterior artery (SCA) and anterior internal carotid artery (AICA) (thin arrow). (C-D) Photograph of a proximal left of PCom (Posterior Communicans) artery grew unruptured saccular aneurysm (bold arrow); left PCom artery (dotted arrow). (E-F) Bilateral vertebral artery hypoplasia is shown in this photograph.*

After careful consideration of the local anatomy, the patient was brought to the angiography suite and we performed endovascular coiling procedure at the unruptured saccular aneurysm on the right side of PCom artery, to block blood flow into an aneurysm (Figure 2). In order to maintain blood flow to posterior circulation system (vertebrobasilar), we did not perform an endovascular coiling procedure on the aneurysm of left PCom artery because PCom artery grew directly from this aneurysm. After the procedure of endovascular coiling, there were no adverse events and further complaints. Three months follow up after endovascular-coiling procedures, the previous symptoms, such as ptosis and ophthalmoplegia significantly resolved (Figure 3).

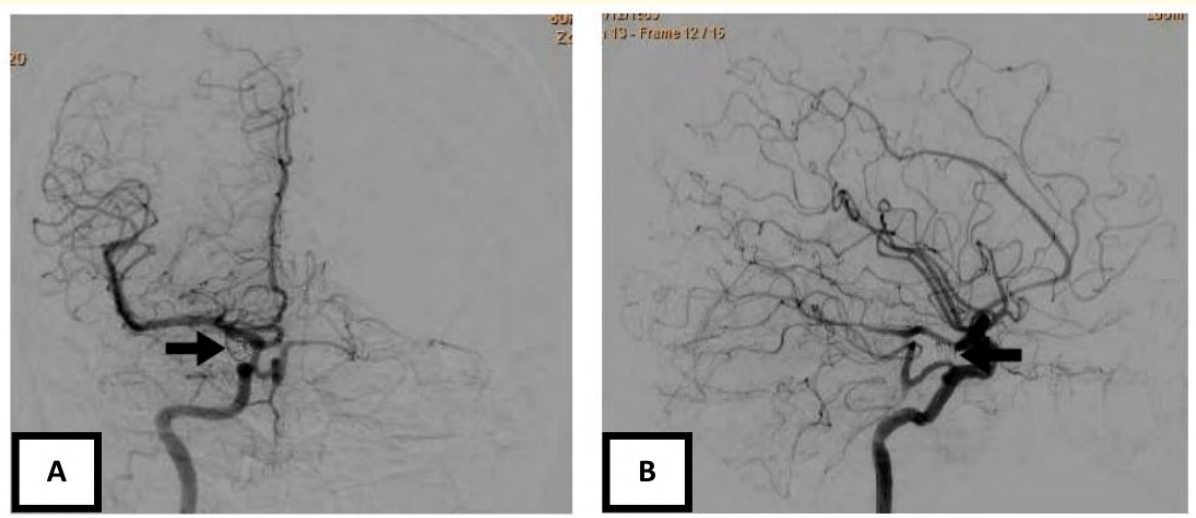

Figure 2: *DSA images after endovascular coiling showed complete obliteration of a right aneurysm (arrow). Photograph from (A) anteroposterior (AP) view of Right ICA and (B)Lateral view of Right ICA.*

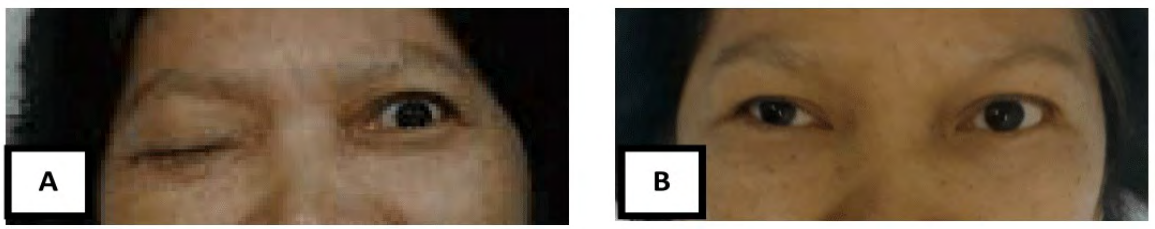

Figure 3: *Condition of patient before and after following endovascular-coiling treatment. (A) Before endovascular coiling, there were right ptosis and right ophthalmoplegia (B) Three months after endovascular coiling, ptosis, and ophthalmoplegia significantly resolved.*

Discussion

Trigeminal artery is the largest fetal anastomosis which connecting CA and BA. It usually disappears on embryonic stage at around 11.5 mm to 14 mm, which makes it become the longest persistent fetal anastomosis of the embryonic period. Hager Padget described the trigeminal arteries as the vascular channels connecting the cavernous portions of the ICA with paired vessels which later forming the BA [1,2].

In the 3 mm embryo stage, the first evidence of the trigeminal artery was described. The dorsal part of aorta has two specific branches on their connection with the first aortic arch; primitive ICA, and PTA. At this stage, trigeminal artery supplies cranial end of primordial hindbrain channel thus proliferates endothelium and gives grow to the veins of the hindbrain and the bilateral longitudinal neural arteries which later becoming BA. In the next 4 - 5 mm of embryonic stage, the trigeminal artery supplies the cranial end of each neural artery [1]. In this stage, an anastomosisis formed, connecting the distal ICA and the corresponding longitudinal neural artery, which sequentially developing into PCom artery. Hence, the pre-segmental arteries and the proatlantal artery will get smaller and finally disappear. With the development of the PCom artery and fusion of the paired longitudinal neural arteries which later forming the BA, these anastomosis will start to get smaller and disappear at particular embryonic stage. If such mechanism didn't work, then the anastomosis between CA and BA will continuously be persistent [3-5].

The PTA has been grouped by Ohshiro and colleagues into two types of classification, a medial type and a lateral type. The medial type of PTA passed through the dorsum sellae and enters the duramater near the clivus. Whereas a lateral type of PTA pass between the sensory root of the fifth nerve and lateral side of the sellae, and running through duramater located in the medial side of Meckel's cave [1,6]. They reported that 13 (59%) were a lateral type and nine (41%) were medial out of 22 cases of PTA detected by autopsy [2].

Based on angiographic, Saltzman classified PTA into three types. In Saltzman type I, PTA runs into the BA at the level that connecting SCA and AICA. Usually, the proximal BA and PCom artery are hypoplastic. The PTA supplies both posterior cerebral artery (PCA) and SCA. In Saltzman type II, the PTA is located right above SCA, which found to enter the BA. The PCom arteries are present and supply the PCA. Combination of types I and II is classified as Saltzman type III. Ali and colleagues further classified the subtypes of Saltzman type III, where PTA grows from the ICA and directly ends on the SCA (as type IIIa), AICA (as type IIIb), or PICA (as type IIIc) without BA being interposition. Thus, based on Saltzman type, the PTA in this case report is classified as Saltzman type II [1,2,4,5,7,8].

PTA bifurcation has a big chance to form an aneurysm although the possibility is far less compared to other bifurcations. The PTA aneurysms could rise on the bifurcation between the segment of cavernous ICA and PTA, on the PTA trunk, or at its junction with BA. PTA aneurysms may present with mass effect in the cavernous sinus, which leads to third or fourth cranial nerve palsy. On the other hand, this case showed unruptured saccular aneurism located at ICA and PCom arterial bifurcation more to postero-inferio-lateral side where the third and fourth cranial nerves run from the posterior side up to anterior one (cavernous sinus), thus giving compression to those nerves and subsequently leading to ptosis and ophthalmoplegia. Such abnormalities finally improved after going through endovascular coiling which stopped the progressivity of aneurism size and even made it smaller, thus the process of entrapment neuropathy in those nerves was reported to get better. The PTA, in this case, was not directly related to the third and fourth cranial nerve palsies, yet it was reported to be a predisposition factor of the aneurism formation for its high haemodynamic from ICA to PCA via the PCom artery. Moreover, the rupture of this type of aneurism can cause an acute subarachnoid haemorrhage, with or without fifth cranial nerve involvement. Balloon-assisted and stent-assisted coiling procedures are the favourable choices of endovascular management in the case of PTA aneurysms [3].

Conclusion

Persistent trigeminal artery is a rare and the most common variant of primitive embryogenic anastomosis that persists and usually associates with any other vascular malformations, such as aneurysm. Angiography is the most reliable method to diagnose and explore any vascular anatomical variants and endovascular-coiling proved as best management for correcting the clinical symptoms.

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