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Case Report

Sequential aneurysms with incidental persistent primitive trigeminal artery: Is this association purely coincidental? A case study and review of the literature in search for a pathobiological mechanism

Sanjeev A. Sreenivasan[1], Neha Agarwal[2], Sudipta Roychowdhury[1], Priyank Khandelwal[3], Anil Nanda[1], Gaurav Gupta[1]

[1]Department of Neurological Surgery, Rutgers - Robert Wood Johnson Medical School, New Jersey, [2]Fetal Centre, Department of Obstetrics and Gynecology, University of Texas McGovern Medical School, Houston, Texas, [3]Department of Neurological Surgery, New Jersey Medical School, RUTGERS, Newark, New Jersey, United States.

E-mail: Sanjeev A. Sreenivasan - sa2034@rwjms.rutgers.edu; Neha Agarwal - nehadas2k@gmail.com; Sudipta Roychowdhury - sroychowdhury@univrad.com; Priyank Khandelwal - pk544@njms.rutgers.edu; Anil Nanda - an651@rwjms.rutgers.edu; *Gaurav Gupta - guptaga@rwjms.rutgers.edu

***Corresponding author:**
Gaurav Gupta,
Department of Neurological Surgery, Rutgers - Robert Wood Johnson Medical School, New Jersey, United States.

guptaga@rwjms.rutgers.edu

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ABSTRACT

Background: The appearance of sequential bilateral aneurysms in patients with persistent primitive trigeminal artery (PTA) is not described in the literature. No clear guidelines on screening and follow-up of patients with incidental PTAs for the detection of associated lesions have been established.

Case Description: A 55-year-old lady presented with occasional headaches. Detailed evaluation showed a left ophthalmic segment internal carotid artery (ICA) aneurysm measuring (11.2 × 5.5) mm. A bovine aortic arch configuration (type III) and a persistent left PTA were identified. A pipeline flex flow diverter was placed and aneurysm was coiled. Follow-up angiography after 6 months showed a completely occluded aneurysm with preserved PTA and a new aneurysm in the right ICA ophthalmic segment measuring 3.5 × 1.5 mm. It was followed up serially with angiography which revealed significant increase in 6 months. The base measured 5.4 mm and two blebs 3.1 mm and 2.5 mm had appeared on the dome. A SUPRASS flow diverter was placed across this aneurysm. Serial follow-up showed complete occlusion of the left aneurysm and significant reduction in filling of the right aneurysm. A review of the literature identified 34 cases of incidental PTAs associated with 50 aneurysms with increased prevalence of anterior circulation aneurysms in patients with incidental PTA.

Conclusion: Sequential angiography of a patient with incidental PTA and an ICA aneurysm shows presence of a new aneurysm in contralateral circulation and its progression in size and morphology. Detailed large-scale studies are needed to assess the impact of incidental PTA on aneurysm development and management.

Keywords: Aneurysm, Flow diverter, Persistent trigeminal artery

INTRODUCTION

The appearance of sequential bilateral aneurysms in patients with persistent primitive trigeminal artery (PTA) is not described in the literature. There is disputing evidence in favor of long-term screening strategies for patients with incidental PTAs and cerebral aneurysms. We conducted a review with the aim to find out whether the associations of PTA with aneurysms are incidental or does it have a pathobiological mechanism? Does specific embryological or genetic mechanism predispose to aneurysms in patient with PTA? Does it have any clinical implications on long-term follow-up? A Google Scholar and PubMed search revealed 3270 articles where 34 studies had 50 aneurysms with incidental PTAs [Table 1]. A separate search among 223 studies to find histology or genetic analysis of PTA was conducted, but did not reveal any positive outcome. We also describe a rare case to highlight the association of incidental PTA.

CASE DETAILS

A 55-year-old lady presented to the clinic with occasional headaches. She did not have family history of cerebral aneurysms. An outside computerized tomography scan showed hyperdensity in the right supraclinoid region. As further evaluation, she underwent diagnostic digital subtraction angiography (DSA).

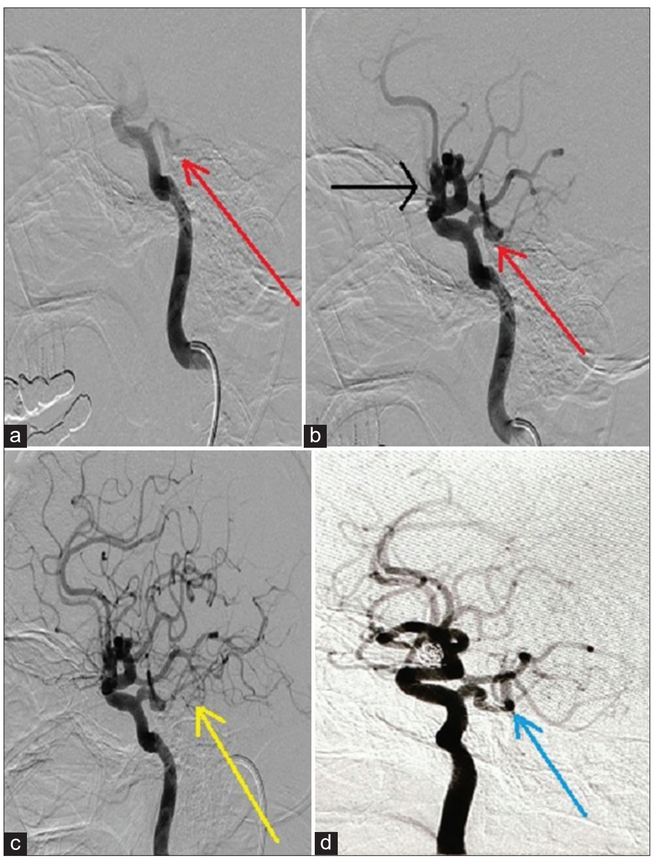

Figure 1: (a) Early filling of PTA (red arrow) from ICA injection (b) filling of the ophthalmic segment ICA aneurysm (black arrow), PTA (red arrow), and upper extent of basilar artery (c) ICA injection image showing filling of posterior circulation (yellow arrow) through the PTA (d) ICA injection image showing filling of PTA with upper extent of basilar artery (blue arrow) and bifurcation into posterior cerebral arteries (postoperative image with coiled aneurysm seen).

A persistent trigeminal artery communicating between cavernous portion of the left internal carotid artery (ICA) and basilar artery was noticed [Figure 1]. The presence of PTA was confirmed on injection of ICA [Figure 1] and vertebral artery injection [Figures 2a-c]. A three-dimensional reconstruction showed the aneurysm of size 11.2 × 5.5 mm along with presence of PTA filing the upper extent of basilar artery and posterior cerebral artery bifurcation [Figures 3a and b]. The aneurysm was saccular in morphology [Figure 3b].

On performing aortic arch injection, the origin of innominate and left common carotid arteries appeared extremely close. This represented a bovine arch configuration. A direct origin of the vertebral artery from the left aortic arch was also noticed (type III aortic arch). A fetal posterior communicating artery (Pcom) was also present. After appropriate counseling and consent, she underwent therapeutic angiography. A 5 mm × 16 mm size pipeline flex flow diverter device was deployed across the aneurysmal ophthalmic ICA segment [Figure 4a] and coils of microsphere 4 mm × 7.5 cm, two target −360 coils: 4 mm × 15 cm and one target helical ultra 3 mm × 8 cm were gradually placed into the aneurysm sac [Figure 4b]. During coil placement, no contrast extravasation or distal thromboembolism was noticed. Contrast stasis within the aneurysm was noticed subsequently.

Figure 2: (a) Subtracted angiography image showing vertebral artery injection filling the PTA (black arrow) and cavernous segment of ICA, (b) sequential angiography image showing filling of ICA ophthalmic segment of ICA and faint filling of aneurysm (red arrow) via the PTA (black arrow), and (c) sequential angiography image showing filling of anterior circulation (black arrow) via PTA, after a vertebral injection.

Table 1: A list of studies with cerebral aneurysms and incidental persistent primitive trigeminal artery.

Study Number	Author	Year	Side	Aneurysm location	Comments
1	Kosnik *et al*.	1977	Bilateral	ICA	
2	Matsuda *et al*.	1979	Right	Posterior communicating artery aneurysm	With subarachnoid hemorrhage
3	Tran-Dinh *et al*.	1984	Right	ICA and anterior communicating artery aneurysm	
4	Matsumura *et al*.	1985	Left	ICA	Polycystic kidney disease, cavum septum pellucidi
5	Sugiyama *et al*.	1987		Anterior communicating artery aneurysm	Surgical clipping
6	Yamanaka *et al*.	1987	Right	ICA	
7	Miyatake *et al*.	1990	Left	Cavernous ICA	
8	Abe *et al*.	1994	Right	Middle cerebral artery	
9	Nakayama *et al*.	1994	Right	Middle cerebral artery	
10	Ishigura *et al*.	1995		Anterior communicating artery aneurysm	
11	De Oliviera *et al*.	1997		Middle cerebral artery	
12	Maeshima *et al*.	1999		(8) Anterior cerebral Artery aneurysms	
13	Baskaya *et al*.	2000	Left	Posterior communicating artery	
14	Ikushima *et al*.	2001		Basilar artery	
15	Ohata *et al*.	2004		Anterior communicating artery	Subarachnoid hemorrhage
16	Behari *et al*.	2004	Right	Posterior communicating artery aneurysm	
17	Schlamann	2006	Right	Posterior cerebral artery	Balloon embolization of aneurysms through PTA
18	Memis *et al*.	2007	Right	Cavernous ICA left side	
19	Sherkat *et al*.	2008	Left	Middle cerebral artery	
20	Aronson *et al*.	2008		Posterior meningeal artery pseudoaneurysm	With subarachnoid hemorrhage
21	Zhang *et al*.	2009	Right	Cavernous ICA	
22	Yamamoto *et al*.	2011	Bilateral	Middle cerebral artery	With subarachnoid hemorrhage
23	Aguiar *et al*.	2011		Basilar artery	With subarachnoid hemorrhage
24	Lei Yan *et al*.	2013		(3) Anterior communicating artery aneurysms	
	Lei Yan *et al*.	2013		(3) Posterior communicating artery aneurysms	
	Lei Yan *et al*.	2013		Middle cerebral artery aneurysm	
25	Alonso-Vanegas *et al*.	2016	Right	Posterior communicating artery	
26	Lam *et al*.	2018	Right	Cavernous ICA aneurysm distal to PTA	
27	Zenteno *et al*.	2018		Anterior communicating artery aneurysm	Hypoplastic vertebral artery
28	Bahar *et al*.	2018		Posterior communicating artery	
29	Zhang *et al*.	2019		Basilar artery	
30	Wan *et al*.	2019		(3) Multiple anterior circulation aneurysms	
31	Kun hou *et al*.	2019		(2) Multiple aneurysms with moyamoya disease	
32	Soylu *et al*.	2019	Left	BA-superior cerebellar artery junction	Endovascular treatment through PTA
33	Bechri *et al*.	2020	Right	Posterior meningeal artery aneurysm	
34	Ito *et al*.	2022	Left	Superior cerebellar artery dissecting aneurysm	With subarachnoid hemorrhage

A follow-up angiogram at 6 months demonstrated complete occlusion of the left ICA aneurysm [Figure 5] and no residual filling or intrastent stenosis was noticed. This angiogram revealed a new second aneurysm arising from the right supraclinoid ICA [Figure 6a]. It measured 3.5 mm × 1.5 mm [Figures 6a and b]. It was decided to follow the patient with serial angiograms. Angiography at 6 months showed good reconstruction of the left ICA with the pipeline flex device. Progression of the right supraclinoid ICA aneurysm in size and morphology was noticed [Figures 7a and b]. Two blebs measuring 3.1 mm and 2.2 mm were noticed on the aneurysm dome and the base of aneurysm measured 5.4 mm. Contralateral persistent trigeminal artery and fetal Pcom artery were noticed intact with good flow.

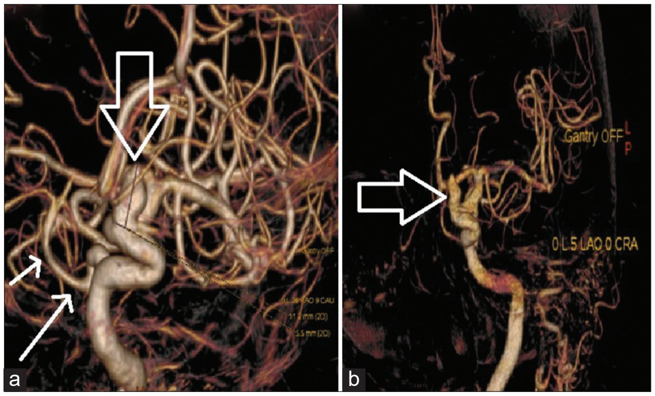

Figure 3: (a) A 3D reconstructed image showing size measurement of the ophthalmic segment ICA aneurysm (hollow white downward arrow) and presence of PTA filling the basilar trunk and bifurcation (solid white arrows) and (b) a 3D reconstructed image showing the saccular morphology of the aneurysm (hollow white arrow).

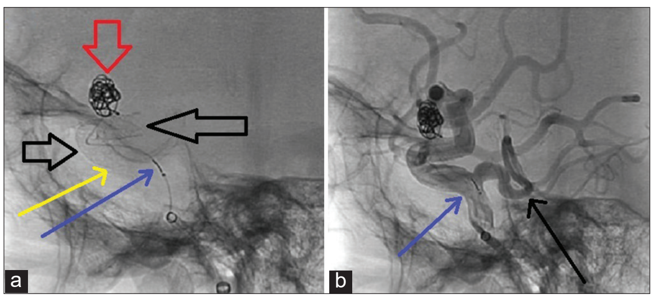

Figure 4: (a) Unsubtracted angiography image showing presence of pipeline flex flow diverter (hollow black arrow) across ICA ophthalmic segment and presence of coils (red arrow) within the aneurysmal space. The pipeline device is carefully deployed so that it stops distal to the origin of PTA (shown with blue arrow toward the black marker of coil). The proximal end of pipeline device is shown with a solid yellow arrow and (b) solid black arrow has been cited in the text of legend as intact PTA draining into upper basilar trunk and posterior circulation. Blue arrow in figure corresponds to proximal coil marker at the level of origin of PTA from ICA.

A single SURPASS flow diverter device measuring 5 mm × 20 mm was placed across the right supraclinoid ICA aneurysm origin [Figure 8 – black arrows]. All the tines of the device were not approximated to the endothelium. Balloon angioplasty was, hence, performed with a 4 mm × 10 mm balloon.

A 6-month follow-up angiography showed that majority of this new aneurysm was thrombosed, with an extremely small residual 0.9 mm aneurysm on the right side ICA [Figures 9a and b]. It was decided to conservatively manage this residual aneurysm with subsequent angiography. Our total follow-up duration for this patient was 18 months with DSA performed after every 6 months.

DISCUSSION

The most common fetal intracranial anastomosis to survive unto adulthood is the persistent PTA.[2] The otic, hypoglossal, and intersegmental arteries rarely persist beyond embryonic stage. In Saltzman type I circulation, the PTA joins distal to anterior inferior cerebellar artery (AICA) and proximal to superior cerebellar artery (SCA). Here, dominant supply to SCA comes from the PTA. In type II, the distal basilar tip is hypoplastic. The posterior cerebral arteries (PCAs) receive blood supply form patent Pcom and PTA supplies the SCA bilaterally. A rare type III variant has a PTA which continues as the AICA directly. [2,5]

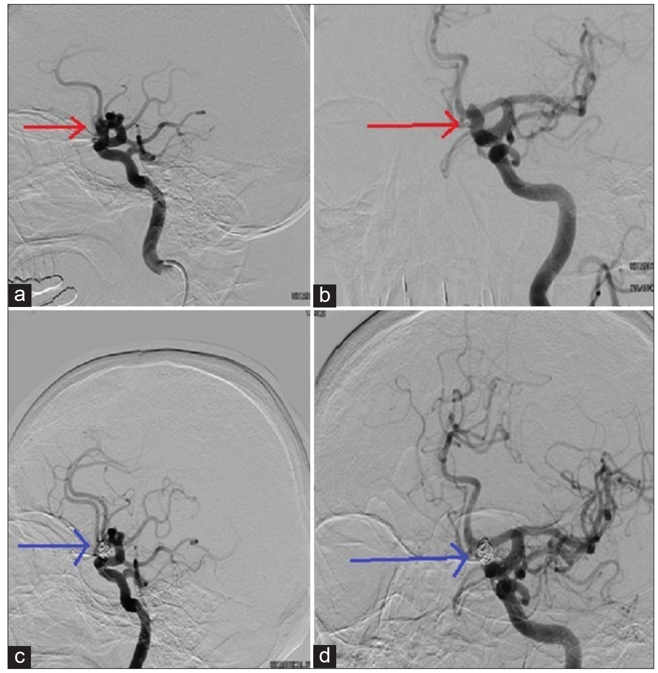

Figure 5: (a) Preoperative lateral angiography image showing the ICA aneurysm (red arrow), (b) preoperative anteroposterior view showing the ICA aneurysm (red arrow), and (c) postoperative lateral angiography image showing coiled aneurysm(blue arrow). A doubt of residual filling seen along the anterior border of aneurysm did arise after this image (d) subsequent anteroposterior image showing complete occlusion of the aneurysm (blue arrow) with no residual filling. The doubt of residual filling on lateral image was clarified as an overshadow of anterior cerebral artery circulation here.

Association of PTA with aneurysms

The presence of aneurysms in patients with PTA is reported around 12–14%.[1] The debate on whether aneurysms are more commonly associated with PTAs than the general population has not been answered (3.7%).[4] The primary aneurysms of the PTA trunk or ICA-PTA junction are rarer (<3–4%).[10] Like any vessel bifurcation, the ICA-PTA junction is prone to develop aneurysms too. A giant thrombosed ICA aneurysm causing hypopituitarism and associated with a PTA has been reported.[15] Maeshima *et al*. reported eight aneurysms of bilateral ACA and MCA.[11] A basilar artery aneurysm associated with PTA was treated with coil embolization with a microcatheter through PTA.[7] Multiple aneurysms in fenestrated MCA and ICA-PTA junction were reported with PTA.[16] A PTA with fetal Pcom

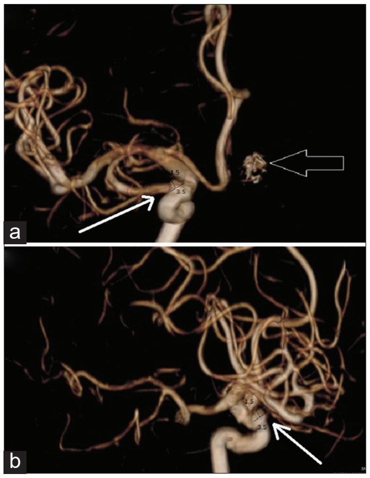

Figure 6: (a) A 3D reconstructed image showing presence of coils (hollow white arrow) and ophthalmic segment ICA aneurysm in the right ICA (solid white arrow) and (b) 3D reconstructed image showing 3.5 × 1.5 mm aneurysm in the right ICA-ophthalmic segment (solid white arrow).

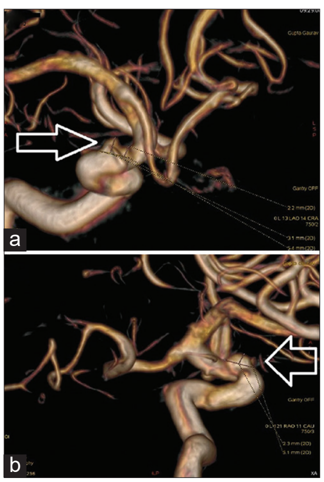

Figure 7: (3D reconstructed images presented) (a) Morphology of the right ICA ophthalmic segment aneurysm (hollow white arrow) seen in follow-up angiography – two blebs seen measuring 2.2 mm and 3.1 mm and maximum base measurement 5.4 mm and (b) appearance of the right ICA aneurysm (white arrow) as seen with base measurement 5.1 mm.

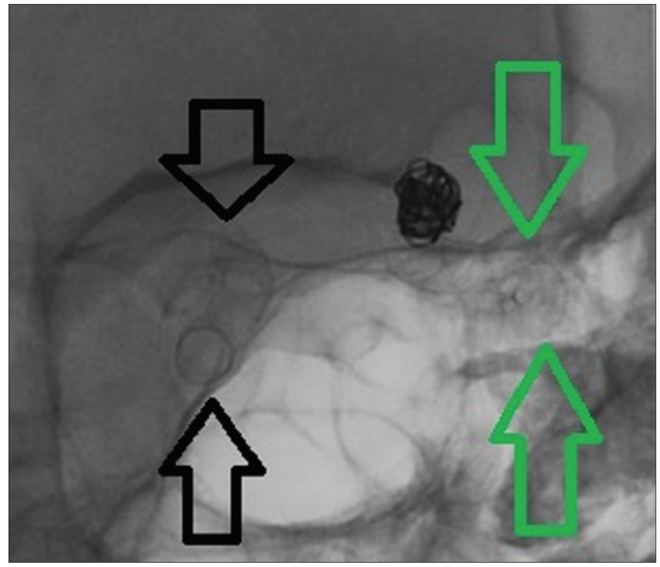

Figure 8: Postoperative follow-up angiography image (unsubtracted) showing right ICA SURPASS flow diverter (hollow black arrows) and left ICA pipeline flex flow diverter (hollow green arrows) along with the presence of coils.

Figure 9: (a) Postoperative 3D reconstructed image showing appearance of the right ICA ophthalmic segment and (b) postoperative 3D reconstructed image showing a small residual aneurysm on ICA ophthalmic segment as seen pointed out by the solid white arrow.

and hypoplastic vertebral artery was known. This patient had a ruptured Pcom aneurysm which was clip repaired.[3]

Simultaneous occurrence of persistent intersegmental artery with PTA and aneurysm on PTA has been reported.[13] Despite these associations, several studies have debated the need of increased surveillance in these patients with incidental PTAs.[6,8]

We believe that the presence of PTA might alter the flow dynamics of circle of Willis, predisposing to aneurysm formation. Other pathogenic mechanisms of aneurysm formation include hypertension and hemodynamic stress. A PTA represents a developmental structural abnormality. The presence of PTA creates additional hemodynamic stress on ICA and BA. The PTA is a vessel situated in lateral parasellar region usually. Rarely, a medial sphenoidal variant (Salas) may be seen. The latter one may be associated with hypopituitarism. The abnormal origin of PTA from ICA and basilar artery predisposes to two regions of hemodynamic stress – one at the ICA posteromedial wall and one at the anterolateral wall of basilar artery. As per Rhoton's theory, the presence of additional branch points in a vessel predisposes to aneurysm formation. The standard anatomy of basilar artery consists of two branch points of PCAs, two for superior cerebellar arteries within 5–8 mm of each other. The presence of another branch point within few millimeters of these will definitely cause added hemodynamic stress. The relative frequency of aneurysms near ICA-PTA junction and anterior circle of Willis were more common than near PTA-BA junction and the posterior circle. Of the 50 aneurysms seen in our literature review, majority were localized to ACA/Acom region, while ICA and PCom aneurysms were equal (eight each), and fewer MCA (6), BA (3), PCA (1), and SCA (2) aneurysms were noticed. This indicates an increased hemodynamic stress in the anterior circulation due to the presence of PTA.

The presence of PTA provides additional anatomic weak spots/pressure points: anatomical branch points act as weak pressure points. There are few cadaveric studies which highlight presence on PTA with associated aneurysms. We searched PubMed for 223 articles with key words – PTA, histology, and genetic basis; but none revealed a histological analysis from cadaveric studies. The anatomical composition of a PTA will suggest the fundamental basis of pathologies associated with it. It would be worthwhile to study the histology of PTA vessel wall – the tunica intima, media, and externa. A genetic analysis of the vessel wall would highlight the basic difference between a normal intracranial vessel and persistent carotid basilar anastomotic remnant.[14]

The location of aneurysm in relation to PTA is a decisive factor for therapy. An aneurysm which involves the ostium of PTA will need careful handling. Risk of occlusion of PTA with coils during endovascular treatment cannot be negated. Prolapse of coils into the parent artery with occlusion of PTA is another risk of treatment. Retrograde filling of aneurysm through a PTA needs to be considered. Simple ligation of aneurysm (while clipping) may not be sufficient when the PTA supplies blood to the aneurysm too.

The physiological significance of PTA depends on the type of circulation. In a fetal type, basilar system and PCAs are supplied by PTA. Occlusion of PTA can be hazardous here. In adult-type circulation, the PTA plays a rudimentary role. Risk of significant clinical consequences is low in this type. In our patient, the flow diverter stent encompassed the origin of PTA. While deploying the stent, it was essential to ensure complete apposition of the stent with endothelium. Precarious density between the endothelium and stent would predispose to thromboembolism of PTA and, hence, ischemia of brainstem perforators. Symptoms of posterior circulation stroke such as distal vertebrobasilar insufficiency will be seen in case of a fetal-type PTA thrombosis.

Association of PTA with stroke

In patients with carotid stenosis and incidental PTAs, posterior circulation ischemic stroke has been reported. PTA may also act as a conduit for superselective catheterization in angiography for stroke.

Association of PTA with trigeminal neuralgia

Symptoms of trigeminal neuralgia have been attributed to a PTA in some patients. Surgical manipulation does provide relief from neuralgic symptoms

Other associations

Several case reports documenting association of PTA with moyamoya disease, aneurysms, hemangiomas, and brain tumors such as medulloblastoma and hemangioblastomas, and other pathologies like arteriovenous malformations have been published.[12,9] PTA has been used a conduit for passing endovascular catheter for therapy of aneurysms in nearby circulation.[7,17]

Need of surveillance

An important component of the management of patients with incidentally detected PTA is regular periodic follow-up angiography imaging. Appearance of new aneurysms on the PTA trunk, ICA-PTA junction, or BA-PTA junction or anywhere else in the circulation needs to be identified. In our patient with treatment history for two unruptured aneurysms, certain important features were traced on the follow-up angiography. This included detection of new aneurysms, patency of parent vessel with stent *in situ*, development of in-stent stenosis, thromboembolic occlusion of the PTA/ophthalmic artery/Pcom artery, or other perforator vessels.

CONCLUSION

Persistence of carotid vertebral anastomosis has been associated with aneurysms in various locations. Histological basis of aneurysm formations remains to be seen. Although papers documenting equivocal presence of aneurysms in the patients with incidental PTA have been published, we report a case where patient develops progression of a newly diagnosed aneurysm. This highlights the heterogeneous nature of aneurysms with PTA as an associated factor. The role of active angiographic surveillance in patients with PTA cannot be undermined. Large-scale studies are needed to clarify the role of screening angiography in this subset of patients.

Consent to participate and publish material

Obtained in written from patient and relatives.

Availability of data and material

Available.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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