



Medial type persistent trigeminal artery associated with a saccular aneurysm at its trunk

Akira Uchino¹ · Hitoshi Ohno¹ · Ryushi Kondo¹ · Shoichiro Ishihara¹

Received: 2 August 2020 / Accepted: 11 December 2020 / Published online: 11 January 2021
© The Author(s), under exclusive licence to Springer-Verlag France SAS part of Springer Nature 2021

Abstract

There are four types of fetal anastomosis between the carotid and vertebrobasilar arteries at 5 weeks gestation; from caudal to cranial position, these involve the proatlantal intersegmental, hypoglossal, otic, and trigeminal arteries. Excluding otic artery, these arteries may persist rarely. Persistent trigeminal artery (PTA) is the most common carotid-vertebrobasilar anastomosis, and the medial type (intrasellar) PTA is quite rare, accounting for approximately 10% of all PTA cases. An aneurysm is occasionally found at the origin of the PTA. Rarely, an aneurysm arises at the trunk of the PTA. Using magnetic resonance angiography, we identified a case of medial type PTA with an unruptured saccular aneurysm at its trunk.

Keywords Cerebral aneurysm · Cerebral arterial variation · Magnetic resonance angiography · Medial type · Persistent trigeminal artery

Introduction

According to Padgett [6], there are four types of fetal anastomosis between the carotid and vertebrobasilar arteries at 5 weeks gestation; from caudal to cranial position, these involve the proatlantal intersegmental, hypoglossal, otic, and trigeminal arteries. The otic artery never persists, and other arteries may persist rarely. An anastomotic artery between the cavernous segment of the internal carotid artery (ICA) and the midportion of the basilar artery (BA) is occasionally incidentally found on magnetic resonance angiography (MRA). This is called persistent trigeminal artery (PTA), the most cephalad and most common type of carotid-vertebrobasilar anastomosis [10]. There are two types: lateral (usual) type and medial (intrasellar) type [7], with the latter being quite rare, accounting for approximately 10% of PTAs [2, 11].

An aneurysm rarely arises at the junction between the ICA and the PTA, and an aneurysm can sometimes be found at the trunk of the PTA [1, 3, 4] or PTA-BA junction [9]. Using MRA, we identified a case of an unruptured saccular

aneurysm occurring at the trunk of the medial type PTA, an extremely rare association [1, 3].

Case report

A 73-year-old woman with aortic arch aneurysm underwent cranial magnetic resonance imaging (MRI) and MRA using a 3-T scanner (Magnetom Skyra, Siemens Medical System, Erlangen, Germany) for the evaluation of cerebrovascular diseases, instead of no significant neurological symptoms. MRA was obtained using a standard three-dimensional time-of-flight technique. The imaging parameters were a flip angle of 18°, repetition time of 21.0 s, echo time of 3.69 s, and slice thickness of 0.8 mm.

MRI revealed no significant abnormality except for multiple tiny ischemic foci in the bilateral cerebral hemispheres (not shown). MRA showed a large anomalous artery arising from the cavernous segment of the right ICA. It took a medial course and turned posteriorly. Finally, it anastomosed with the mid portion of the BA, indicative of medial type PTA. At the angulation point of the PTA trunk, a saccular aneurysm was found. Another saccular aneurysm was seen at the distal cavernous segment of the right ICA (Figs. 1, 2). The proximal BA was hypoplastic. The right posterior cerebral artery (PCA) was supplied by the ICA, indicative of

✉ Akira Uchino
auchino0528@gmail.com

¹ Saitama Sekishinkai Hospital, Sayama, Japan

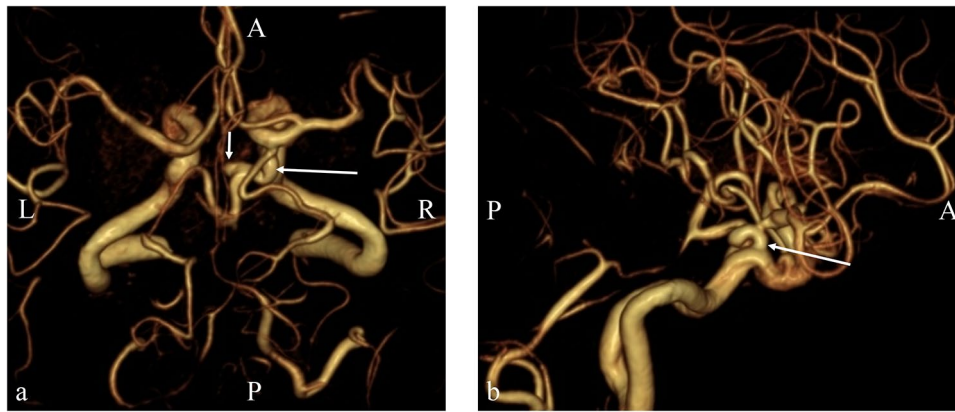


Fig. 1 Supero-inferior (a) and right lateral (b) projections of volume rendering (VR) image of magnetic resonance angiography (MRA) show a large anomalous artery arising from the cavernous segment of the right internal carotid artery (ICA) (long arrows). “Tau sign” is seen in the lateral projection. The artery takes a medial course and

turns posteriorly, finally anastomosing with the basilar artery (BA), which is indicative of medial type (intrasellar) persistent trigeminal artery (PTA). At the turning point of the PTA trunk, a saccular aneurysm is seen (short arrow)

a fetal origin of the PCA. Thus, the PTA of our patient was classified as Salzman’s type 3 [8].

The two aneurysms of this patient were not treated by coil embolization because of the aortic arch aneurysm and lack of ant symptoms related to the aneurysms. Her clinical course was uneventful for over 1 year.

Discussion

There are four types of fetal anastomosis between the carotid and vertebro-basilar arteries (VBA): from the caudal to cranial position, these involve the proatlantal intersegmental, hypoglossal, otic, and trigeminal arteries. Excluding the otic artery, these arteries may persist rarely, with each type manifesting several variations [10]. The PTA is the most common types of anastomosis. According to a meta-analysis by Brzegowy et al. [2], its overall prevalence was 0.3%. Traditionally, Salzman’s classification [8] was used for PTAs. He classified PTAs according to their relationship with the posterior communicating artery (PCoA). Type 1 is absent PCoA bilaterally, Type 2 is absent the P1 segment of the PCA (called fetal origin PCA) bilaterally, and Type 3 is ipsilateral fetal origin of PCA. Thus, our patient is regarded as having Type 3 PTA. This classification may be useful for the hemodynamic evaluation of the VBA, including blood supply via the PTA. However, the infra- and supra-tentorial arteries adopt different routes of embryological development. The PTA is an infratentorial artery, while the PCoA is supratentorial artery. Thus, the PTA and PCoA have no developmental relationship with each other. We, therefore, believe that the Salzman’s classification [8] has no meaning [11].

Reviewing several anatomic reports concerning PTAs, Salas et al. [7] classified the PTA into two types according to relationship to the abducens nerve, lateral and medial types. The lateral type of the PTA originates from the posterolateral aspect of the posterior bend of the cavernous segment of the ICA, and it crosses underneath and distorts the abducens nerve, continuing between the abducens and trigeminal nerves. The medial type PTA originates from the posteromedial aspect of the posterior bend of the cavernous segment of the ICA and pierces the clival dura at the dorsum sellae. Thus, the medial type is also called intrasellar PTA, and it is dangerous to encounter during transsphenoidal pituitary surgery [5]. These two types of PTAs are suggested to have different routes of embryonic development. The medial type is rare, accounting for only roughly 10% of PTAs [2, 11]. The medial type PTA arises from a slightly more distal portion of the ICA than the lateral type does [10].

An aneurysm can be seen at the ICA-PTA junction, less frequently at its trunk [1, 3, 4], and extremely rarely at the PTA-BA junction [9]. According to a literature review by Kai et al. [4], 18 PTA aneurysms were detected at the ICA-PTA junction, 12 at the PTA trunk, and 3 at the PTA-BA junction. The majority of PTA aneurysms are saccular type, but fusiform aneurysm has also been reported. PTA aneurysm may present with mass effect in the cavernous sinus, such as abducens nerve palsy or trigeminal neuralgia. If a PTA aneurysm ruptures, subarachnoid hemorrhage or carotid-cavernous fistula occurs, depending on whether it is located on the intracavernous or extracavernous portion of the PTA. According to the review of PTA aneurysm by Diana et al. [3], medial type PTA aneurysm is only seen in 1 out of 40 cases.

Our patient also had another aneurysm at the distal cavernous segment of the ipsilateral ICA. The frequency of

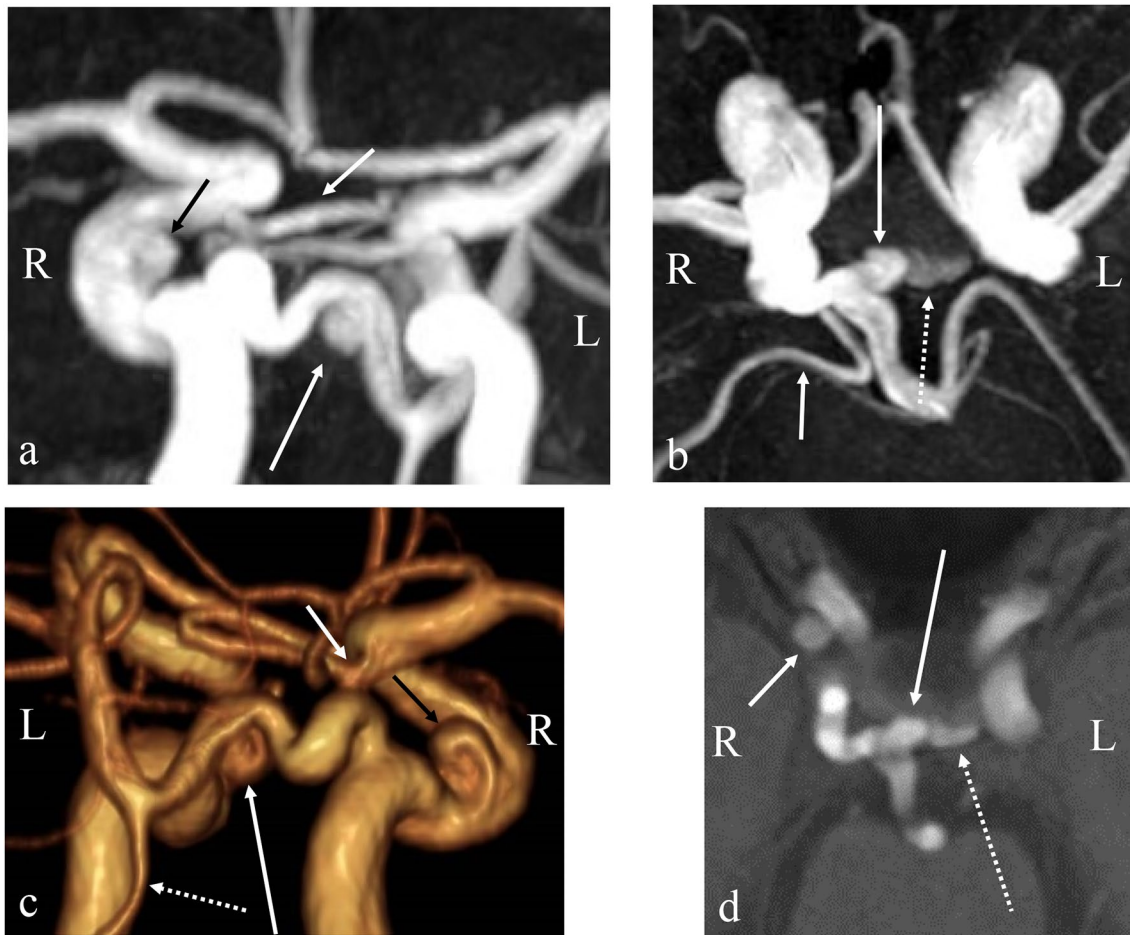


Fig. 2 Left anterior oblique (**a**) and infero-superior (**b**) projections of MRA show a large anomalous artery arising from the cavernous segment of the right ICA. The artery takes a medial course and turns posteriorly, finally anastomosing with the BA, which is indicative of medial type PTA. At the turning point of the PTA trunk, a saccular aneurysm is seen (*long arrows*). The right posterior cerebral artery (PCA) arises from the ICA, which is indicative of fetal type PCA (*short white arrows*). Thus, this is classified as Salzman's type 3 PTA. There is another saccular aneurysm at the distal cavernous segment of the right ICA (**a**, *short black arrow*). The *dotted arrow* (**b**) indicates the posterior lobe of the pituitary gland. **c** Right posterior oblique projection of the VR image of MRA shows the anatomical details

of the anomalous artery and its surrounding arterial structures. The *long arrow* indicates saccular aneurysm of the PTA trunk. The *short black arrow* indicates saccular aneurysm of the distal cavernous segment of the right ICA. The *short white arrow* indicates the origin of the fetal type right PCA. The *dotted arrow* indicates hypoplastic BA. **d** Reformatted source image of MRA at 5 mm thickness shows the anatomical relationship between the medial type PTA and aneurysms. The long arrow indicates saccular aneurysm of the PTA trunk. The short arrow indicates saccular aneurysm of the distal cavernous segment of the right ICA. The dotted arrow indicates the posterior lobe of the pituitary gland

intracranial aneurysms in other locations coexisting with a PTA was reported to be 4.2% using MRA, which is similar to that in the general population (3.7%). Thus, other intracranial aneurysms seem to have no association with the PTA. The previously reported higher frequency of cerebral aneurysms in patients with a PTA might be related to selection bias, as catheter angiographic studies are usually applied for evaluating symptomatic aneurysms.

Ruptured PTA aneurysms should be treated by coil embolization, with a balloon-assisted technique [4, 9], or with a dual stent-assisted technique. Recently, the flow-diverter stent has been used for aneurysm treatment. If preservation of the PTA

proves difficult, the PTA trunk may be occluded by coils [3]. Symptomatic unruptured aneurysms should also be treated. The aneurysms in our patient were unruptured, and there were no related symptoms noted. In addition, she was 73 years old and had an aortic arch aneurysm. Therefore, she was treated conservatively.

Conclusions

We reported a case of medial type (intrasellar) PTA associated with an unruptured saccular aneurysm at its trunk, an extremely rare association. Only a few cases have been reported previously. The Salas classification of the PTA (lateral and medial types) is more useful than Salzman's classification, including its modifications.

Author contributions AU carried out the study design and drafted the manuscript. All authors reviewed the manuscript critically, and have read and approved the final manuscript.

Compliance with ethical standards

Conflict of interest The authors declare that we have no conflict of interest.

References

- Bai M, Guo Q, Sun Y (2014) Rare saccular aneurysm in a medial type persistent trigeminal artery trunk and literature review. *Surg Radiol Anat* 36:299–302
- Brzegowy K, Pekala PA, Zarzecki MP, Pekala JR, Roy J, Aziz HM, Tubbs RS, Walocha JA, Tomaszewski KA, Mikos M (2020) Prevalence and clinical implications of the primitive trigeminal artery and its variants: a meta-analysis. *World Neurosurg* 133:e401–e411
- Diana F, Mangiafico S, Valente V, Wlclerk A, Grillea G, Colonnese C, Bartolo M (2019) Persistent trigeminal artery aneurysms: case report and systematic review. *J Neurointerv Surg* 11:1261–1265
- Kai Y, Ohmori Y, Watanabe M, Morioka M, Hirano T, Kawano T, Sakurama T, Miura A, Kuratsu J (2011) Coil embolization of an aneurysm located at the trunk of the persistent primitive trigeminal artery. *Neurol Med Chir (Tokyo)* 51:361–364
- Lee KS, Kelly DL, Kelly DL Jr (1989) Intrasellar persistent trigeminal artery associated with a pituitary adenoma. Case report. *J Neurosurg* 70:271–273
- Padget DH (1948) The development of the cranial arteries in the human embryo. *Contrib Embryol* 32:205–261
- Salas E, Ziyal IM, Sekhar LN, Wright DC (1998) Persistent trigeminal artery: an anatomic study. *Neurosurgery* 43:557–561
- Saltzman GF (1959) Patent primitive trigeminal artery studied by cerebral angiography. *Acta Radiol* 51:329–336
- Shah KA, Katz JM (2020) Ruptured persistent trigeminal artery-basilar artery junction aneurysm: case report and review of literature. *World Neurosurg* 133:159–162
- Uchino A (2019) Carotid-vertebrobasilar anastomosis: magnetic resonance and computed tomographic angiographic demonstration. *Jpn J Radiol* 37:565–578
- Uchino A, Saito N, Okada Y, Kozawa E, Mizukoshi W, Inoue K, Takahashi M (2012) Persistent trigeminal artery and its variants on MR angiography. *Surg Radiol Anat* 34:271–276

Publisher's Note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.