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Case Report

Ruptured cerebral aneurysm associated with a persistent primitive trigeminal artery variant

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Abstract

Background: Primitive trigeminal artery variants (PTAVs) are one of the rare persistent fetal anastomoses between the carotid and vertebrobasilar circulations. They originate from the internal carotid artery and join one of the cerebellar arteries instead of the basilar artery.

Case Description: We present an 82-year-old woman with subarachnoid hemorrhage due to a ruptured aneurysm originating at a PTAV. Three-dimensional computed tomography angiogram and cerebral angiography revealed bilateral PTAV and two aneurysms originating at the left PTAV. The proximal and distal aneurysms were saccular and fusiform, respectively. She underwent surgical treatment and her postoperative course was uneventful.

Conclusion: Our case demonstrates that extremely rare cerebral aneurysms associated with PTAV can be addressed successfully by surgical intervention.

Key Words: Fusiform, intracranial aneurysm, persistent primitive trigeminal artery variant, saccular, subarachnoid hemorrhage, surgical treatment

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INTRODUCTION

The primitive trigeminal artery (PTA) originates from the internal carotid artery and joins the basilar artery (BA). It is the most common carotid basilar anastomosis, whose incidence is reported to be between 0.1% and 0.6% of individuals.[5,11] PTA variants (PTAVs) originate at the internal carotid artery and join one of the cerebellar arteries instead of the BA.[5] We report an extremely rare case of subarachnoid hemorrhage (SAH) associated with multiple cerebral aneurysms on a PTAV; one aneurysm was saccular and the other was fusiform. Both were treated successfully by surgical intervention.

CASE REPORT

An 82-year-old woman was admitted to the emergency department of a hospital with sudden-onset severe headache. Her past medical history included hypertension and hyperlipidemia, and she had undergone surgery 1 year earlier after SAH due to the rupture of a saccular aneurysm originating at the left internal carotid-posterior communicating artery. Postoperatively, she manifested no neurological disorders and she reported for regular follow-up of her unruptured intracranial aneurysms. Cranial computed tomography (CT) performed at admission revealed SAH (Hunt and Kosnik grade 2, WFNS grade 1,

Fisher group 3) [Figure 1]. She was introduced to our hospital 4 days post onset. Three-dimensional CT angiogram (3D-CTA) and cerebral angiography showed bilateral PTAV and two aneurysms originating at the left side of the vessel [Figures 2, 3]. Comparison with the earlier studies showed that the size of the aneurysms had not changed and we were unable to identify the ruptured aneurysm. The patient manifested cerebral vasospasm and was placed under observation. Her condition remained good and she underwent surgery via a left lateral suboccipital approach on the 18th day post-SAH. The left PTAV penetrated the isolated dural foramen below Meckel's cave and lateral to the dorsum sellae [Figure 4]. Both aneurysms were visualized; one was saccular type at the proximal side and the other was fusiform type at the distal side. We concluded that the saccular aneurysm had ruptured. It was clipped and the fusiform aneurysm was clipped and wrapped [Figure 5]. On the 33rd day post-SAH, we placed a ventriculo-peritoneal shunt for hydrocephalus. Subsequently, her conscious level gradually improved and she was moved to another hospital for rehabilitation.

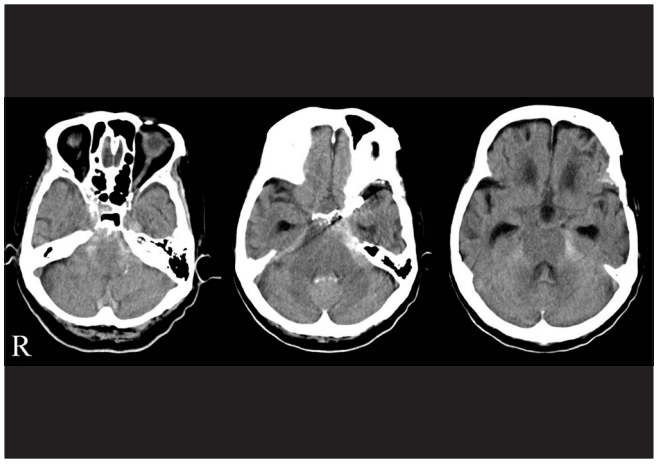

Figure 1: Cranial computed tomography performed on admission demonstrates diffuse subarachnoid hemorrhage in the basal cistern

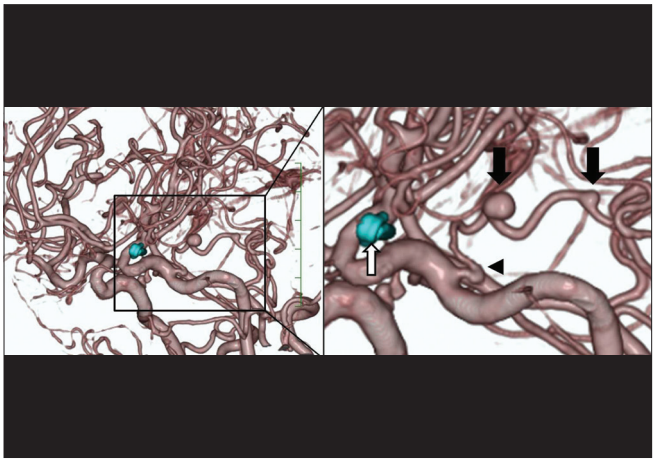

Figure 2: Three-dimensional computed tomography angiogram revealing two aneurysms (arrow) on the left persistent trigeminal artery variant, (arrow head). Note the clip from the earlier procedure to address a previously ruptured left internal carotid-posterior communicating artery aneurysm (open arrow)

DISCUSSION

We addressed a rare case of multiple cerebral aneurysms on a PTAV by surgical intervention.

PTAVs originate at the cavernous or pre-cavernous portion of the internal carotid artery and course along the trigeminal nerve without joining the BA and directly supply the cerebellum, terminating as the cerebellar artery.[5] They are classified as lateral or medial depending on their course. Lateral type runs lateral to the dorsum sella, and medial type runs middle course through or over the dorsum sella.[11] In our case, the left PTAV penetrated the isolated dural foramen below Meckel's cave, consequently it was classified as a lateral PTAV.

Many cases of PTAV were reported to be associated with intracranial aneurysms. The high prevalence of intracranial aneurysms on a PTAV has been explained by the presence of structural defects in the wall of the cerebral arteries.[11,13] Others reported that the incidence of intracranial aneurysms associated with PTAV was 4.0%, similar to the prevalence of these aneurysms in the general population. This study involved 16,415 patients who underwent magnetic resonance imaging (MRI) study at a single institution.[11] Although further investigations are needed, we speculate that the incidence of cerebral aneurysms on a PTAV may be similar to the rate of aneurysms located at other sites.

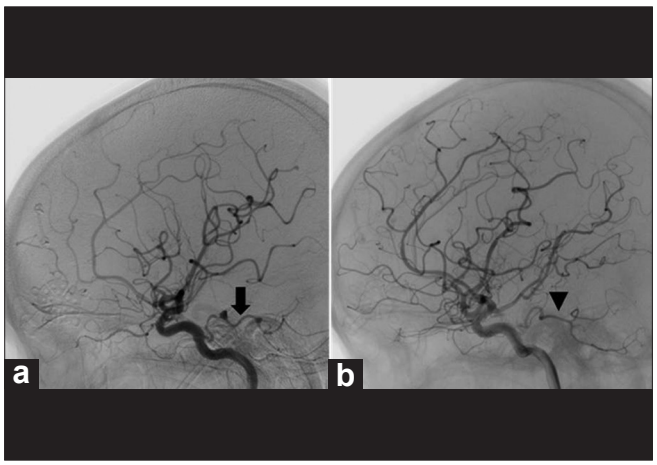

Figure 3: (a) Left internal carotid angiogram showing that the left PTAV supplies the ipsilateral anterior inferior cerebellar artery region (arrow). Note the two aneurysms originating from the vessel. (b) Right internal carotid angiogram showing that the left persistent trigeminal artery variant, supplies the ipsilateral anterior inferior cerebellar artery and the posterior inferior cerebellar artery region (arrow head)

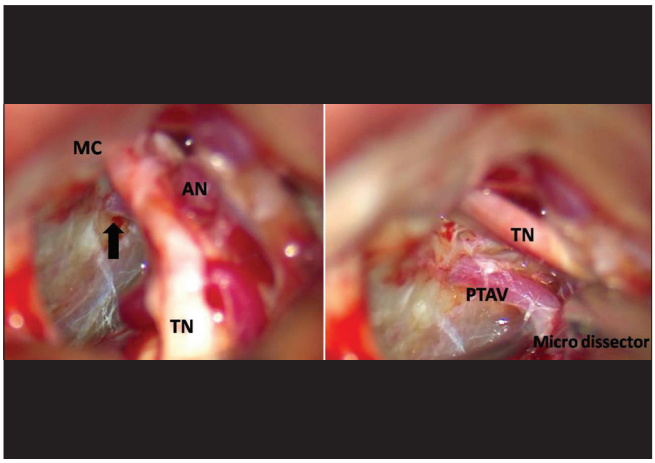

Figure 4: Image obtained before clipping and wrapping the aneurysm. The left persistent trigeminal artery variant, penetrated through the isolated dural foramen below Meckel's cave and lateral to the dorsum sella (arrow). AN: aneurysm, TN: trigeminal nerve, MC: Meckel's cave

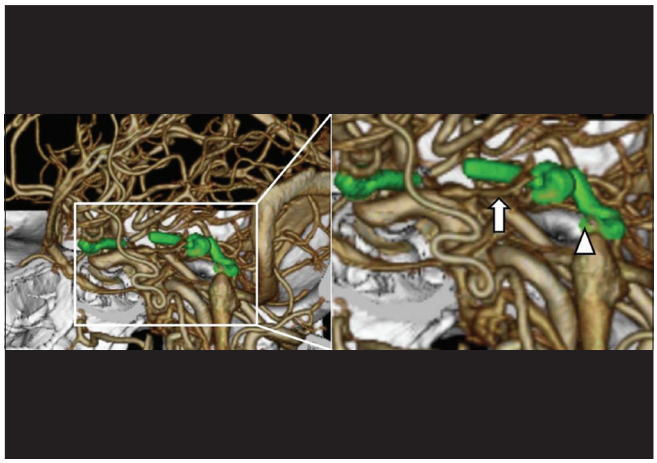

Figure 5: Three-dimensional computed tomography angiogram obtained after surgical treatment. The saccular aneurysm was clipped (arrow) and the fusiform aneurysm was clipped and wrapped (arrow head)

Table 1: Cases of Persistent trigeminal artery variant aneurysms

Case No.	Author	Year	Age (years)	Sex	Side	Aneurysm site	Aneurysm type	Onset and remarks
1	Matsuda *et al.*	1979	32	F	Rt	P-com, ICA-PTAV	Undocumented	Hemorrhage, AVM, multiple AN
2	Watanabe *et al.*	1988	67	F	Rt	ICA-PTAV	Fusiform	Hemorrhage, multiple AN
3	Nioka *et al.*	1993	52	F	Lt	ICA-PTAV	Saccular	Hemorrhage, multiple AN
4	Hayashi *et al.*	1994	47	F	Rt	ICA-PTAV	Saccular	Hemorrhage
5	Hanabusa *et al.*	2000	71	F	Lt	PTAV	Saccular	Hemorrhage, two segments of the left PTAV
6	Nishio *et al.*	2001	69	F	Rt	ICA-PTAV	Saccular	Diplopia
7	Shin *et al.*	2005	40	M	Rt	ICA-PTAV	Aneurysmal dilatation	Hemorrhage, CCF
8	Yang *et al.*	2010	48	M	Rt	PTAV	Fusiform	Hemorrhage, hypoplastic AICA, VA
9	Present case	2011	82	F	Bilateral	MCA, PTAV	Saccular and fusiform	Hemorrhage, multiple AN

M: Male, F: Female, P-com: Posterior communicating artery, ICA: Internal carotid artery, PTAV: Persistent trigeminal artery variant, MCA: Middle cerebral artery, AVM: Cerebral arteriovenous malformation, AN: Aneurysm, CCF: Carotid cavernous fistula, VA: Vertebral artery, AICA: Anterior inferior cerebellar artery, Rt: Right, Lt: Left

To date, nine patients with cerebral aneurysms on a PTAV, including ours, have been documented [Table 1].[2,3,7,9,10,12,14,15] The aneurysms originated at the junction of the internal carotid artery in six cases; three patients presented with distal aneurysms. Our patient presented with bilateral PTAVs and two different types of distal aneurysms on the left side. Although a few cases of different types of aneurysms originating at the same vessel other than a PTAV have been reported, the aneurysms were secondary to radiation therapy or vascular malformation.[6,8] Most of the previously reported aneurysms on a PTAV were saccular; only three were fusiform. The developmental origin of saccular aneurysms has been attributed to a defect in the elastic and muscular coats of blood vessels; fusiform aneurysms are thought to be associated with atherosclerotic processes.[1] We posit that hemodynamic stress on the PTAV induced the saccular aneurysm, same as the other parts of the aneurysms, and the fusiform aneurysms were a consequence of age-related atherosclerotic changes in this case.

The reported interventions for PTA/PTAV aneurysms are endovascular embolization and surgical intervention.[2,3,4,9,10,12,14,15] Our patient underwent direct surgical intervention because it was not clear which of her aneurysms had ruptured and we thought that both the aneurysms required treatment. The subtemporal approach might be useful to address proximal aneurysms in such situations although in cases such as ours we recommend the lateral suboccipital approach to treat both the aneurysms in a single surgical procedure.[4]

CONCLUSION

We presented an extremely rare case of SAH associated with multiple cerebral aneurysms on a PTAV that was treated successfully by direct surgery. Additional studies are needed to clarify the features of PTAV and the relationship between PTAV and aneurysms.

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