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Case Report

Parent Artery Occlusion for Symptomatic Large Internal Carotid Artery Aneurysm with Primitive Trigeminal Artery Variant: A Case Report

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Objective: To report a case of symptomatic large cerebral aneurysm of the internal carotid artery (ICA), associated with a primitive trigeminal artery variant (PTAv), which was treated with a balloon occlusion test (BOT) to evaluate ischemic tolerance.

Case Presentation: A 79-year-old woman was diagnosed with a symptomatic large cerebral aneurysm of the ICA bifurcating the PTAv due to diplopia. After confirming the ischemic tolerance of the perfusion area and PTAv by BOT, we performed ICA parent artery occlusion with selective embolization of the PTAv. Postoperative MRI showed no ischemic lesion and the diplopia was resolved.

Conclusion: ICA parent artery occlusion with PTAv selective embolization after evaluation by BOT is useful in the treatment of large aneurysms.

Keywords primitive trigeminal artery variant, cerebral aneurysm, balloon occlusion test, parent artery occlusion

Introduction

The primitive trigeminal artery (PTA) is the most frequent primitive anastomosis of the internal carotid artery (ICA)–vertebrobasilar artery system, with a frequency of approximately 0.6%. Primitive trigeminal artery variant (PTAv), in which the PTA directly communicates with the cerebellar cortical artery, may be associated with various vascular abnormalities and a balloon occlusion test (BOT) may be necessary before considering a treatment strategy. In the present study, we performed a parent artery occlusion of the ICA with selective embolization of PTAv for a symptomatic ICA aneurysm complicated by PTAv and obtained good results.

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Case Presentation

A 79-year-old woman, experiencing diplopia for a month prior, visited her previous doctor and was referred to our hospital with a cerebral aneurysm in the right ICA visible on MRI. Neurological findings at the time of admission showed right ophthalmoplegia, right abducens nerve palsy, and sensory disturbance of the first branch of the right trigeminal nerve. Head MRI showed a large aneurysm (22 mm) in the cavernous sinus of the right ICA with findings suggestive of partial thrombosis (**Fig. 1A**–**1C**). Cerebral angiography showed a large aneurysm in the cavernous sinus of the right ICA with a PTAv branch proximal neck to the aneurysm (**Fig. 2A**–**2C**). The vessel diameter of the distal ICA was 5.21 mm and that of the proximal ICA was 5.98 × 5.60 mm.

Although a flow diverter stent was considered for the large ICA aneurysm, it was contraindicated because the parent artery diameter was over 5 mm and catheter guidance along the distal ICA to the aneurysm was expected to be difficult. Therefore, we decided to evaluate ischemic tolerance of ICA and PTAv blood supply area by BOT, and

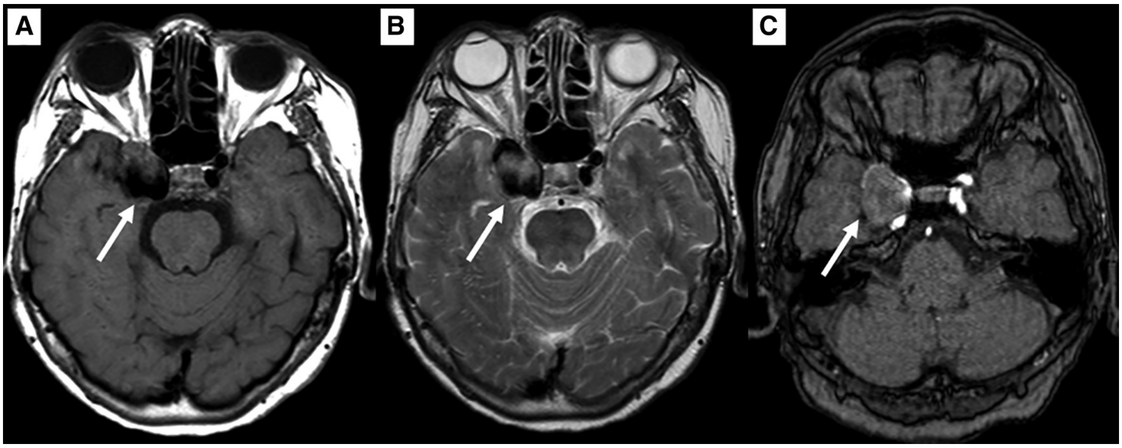

Fig. 1 Initial MRI showing a large aneurysm (arrows) of the right ICA in the cavernous sinus with findings suggestive of partial thrombosis. (**A**) T1-weighted image, (**B**) T2-weighted image, and (**C**) time-of-flight-MRA. ICA: internal carotid artery

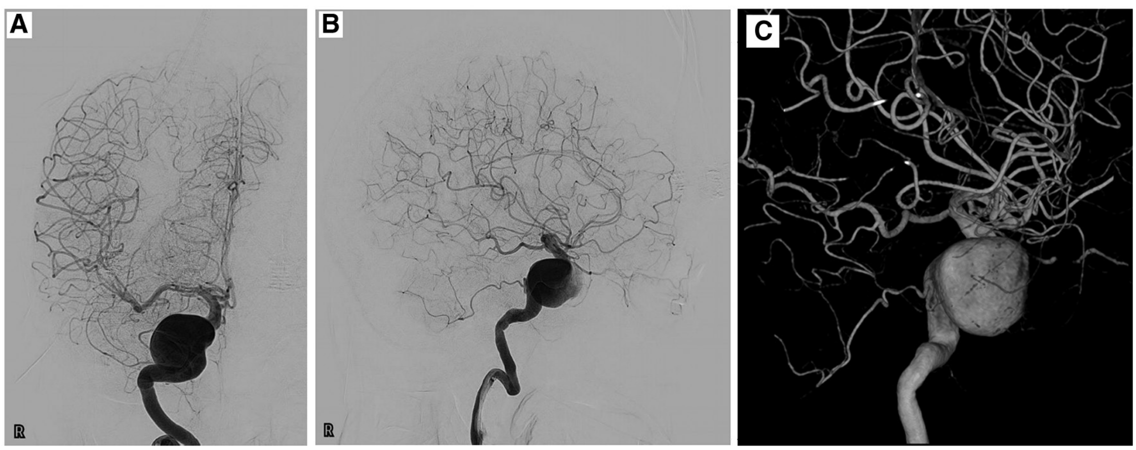

Fig. 2 Pretreatment right internal carotid angiography showing a large aneurysm of the right ICA in the cavernous sinus with a PTAv bifurcated from the aneurysm proximal neck. (**A**) Anterior–posterior view, (**B**) lateral view, and (**C**) 3D rotation angiography. ICA: internal carotid artery; PTAv: primitive trigeminal artery variant

if ischemic tolerance was present, we would perform parent artery occlusion but if not, we would perform trapping with high-flow bypass.

We performed BOT with a 7 × 7 mm SHORYU HR (Kaneka Medics, Osaka, Japan), including the PTAv proximal to the aneurysm, and evaluated ischemic tolerance (**Fig. 3A**). Pan-aortography did not show any left–right difference in the early venous phase (**Fig. 3B**). Right vertebral artery (VA) angiography showed that the right ICA was well perfused through the posterior communicating artery (PcomA), while the basilar artery (BA) was also perfused anterogradely from the right VA (**Fig. 3C**). Near-infrared spectroscopy (NIRS) monitoring (NIRO-200NX; Hamamatsu Photonics, Shizuoka, Japan) also showed no decreases in tissue oxygenation index (TOI). As no neurological symptoms were observed after 20 minutes of balloon occlusion, the patient was judged to be ischemic tolerant. PTAv perfused the right anterior inferior cerebellar artery (AICA) region, but BOT including PTAv showed retrograde blood flow from the VA to right AICA. Furthermore, occlusion was considered possible because there was no hearing impairment or cerebellar symptoms due to PTAv occlusion. The perforating branches of the brainstem from PTAv could not be evaluated, but BOT also showed no obvious brainstem symptoms. As reverse blood flow through the ICA would have fed the PTAv, we decided to selectively embolize the PTAv since the aneurysm might not have been resolved only by occlusion of the parent artery proximal to the ICA.

Under general anesthesia, the right femoral artery was punctured and a 6-Fr FUBUKI 90-cm ST (Asahi Intec, Aichi, Japan) was guided into the origin of the right ICA using a 5-Fr JB2 and 0.035-inch guiding wire. Intraoperatively, heparin was administered and activated clotting time

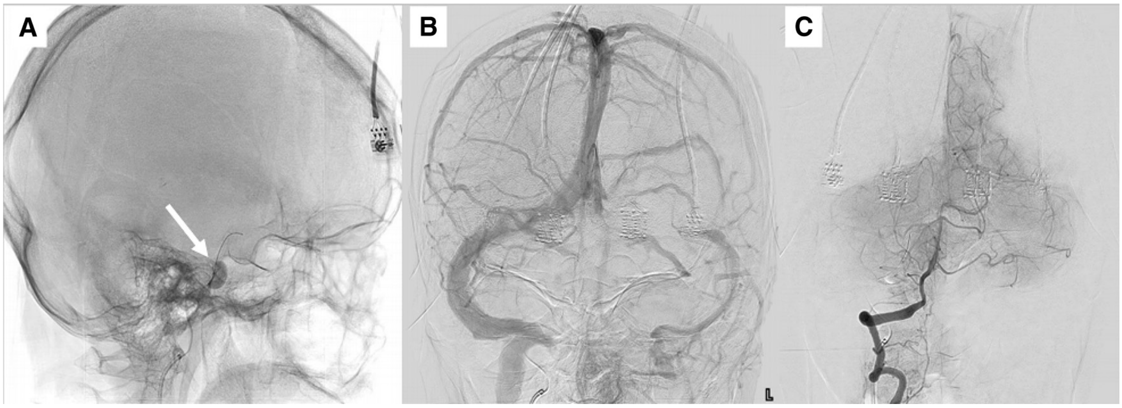

Fig. 3 BOT images. (**A**) Live image of BOT showing balloon (arrow) dilation in the ICA including PTAv. (**B**) Pan-aortography early venous phase showing left–right differences. (**C**) Right vertebral angiography under BOT: Blood flow from the right VA to the BA and retrograde right AICA can be seen. AICA: anterior inferior cerebellar artery; BA: basilar artery; BOT: balloon occlusion test; ICA: internal carotid artery; PTAv: primitive trigeminal artery variant; VA: vertebral artery

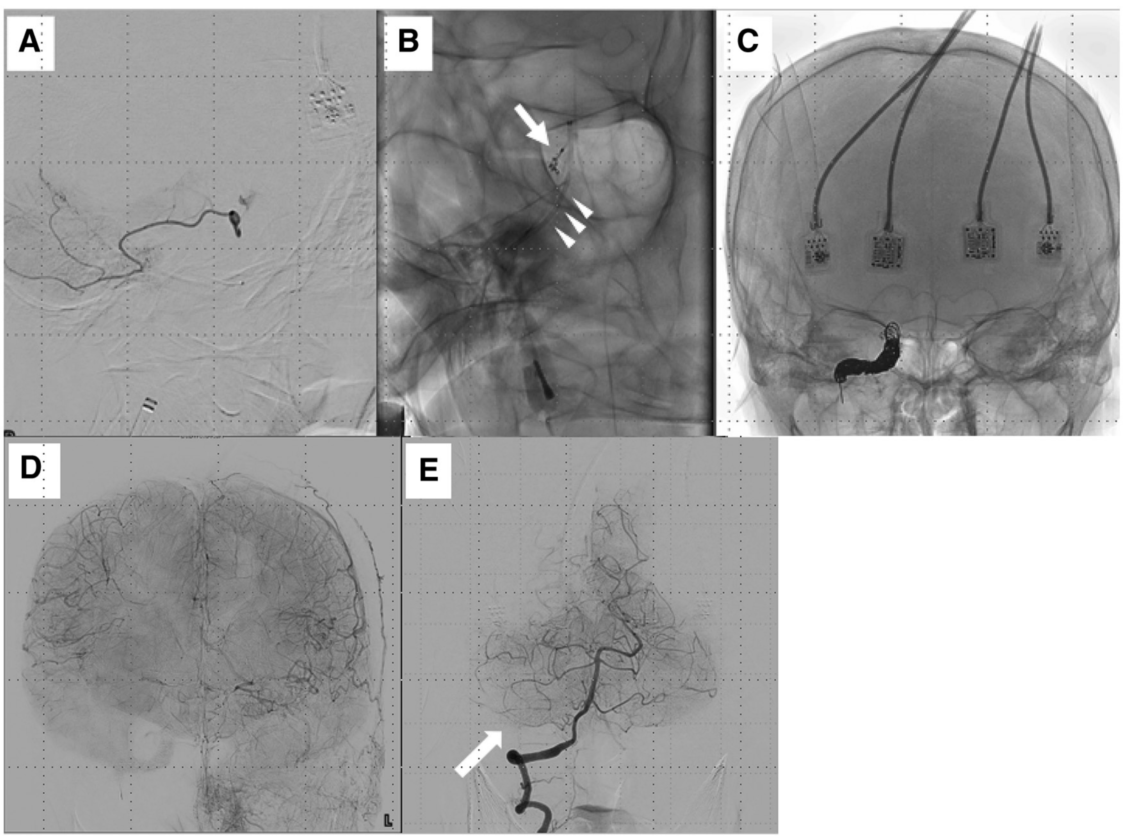

Fig. 4 Images from the coil embolization. (**A**) Superselective angiogram of the PTAv showing blood flow in the right AICA region. (**B**) PTAv embolization with coils (arrow: PTAv embolization site, arrowheads: microcatheter). (**C**) Live image of parent artery occlusion of the right ICA with coils. (**D**) Left internal carotid angiography after occlusion showing good right ICA territory blood flow via the anterior communicating artery. (**E**) Right VA angiography after occlusion showing retrograde blood flow in the AICA territory (or region) (arrow: AICA region perfused with leptomeningeal anastomosis). AICA: anterior inferior cerebellar artery; ICA: internal carotid artery; PTAv: primitive trigeminal artery variant; VA: vertebral artery

was adjusted to 200s–250s. An Excelsior SL10 (Stryker, Fremont, CA, USA) was inserted into the PTAv with a CHIKAI 0.014 inch 200 cm (Asahi Intec). A LANTERN microcatheter (Medicos Hirata, Tokyo, Japan) was then guided to the aneurysmal origin in the ICA and an Axium Prime 3D ES 1 mm × 2 cm coil (Medtronic, Minneapolis, MN, USA) was inserted through the SL10 to occlude the PTAv (**Fig. 4A** and **4B**). Subsequently, a total of eight

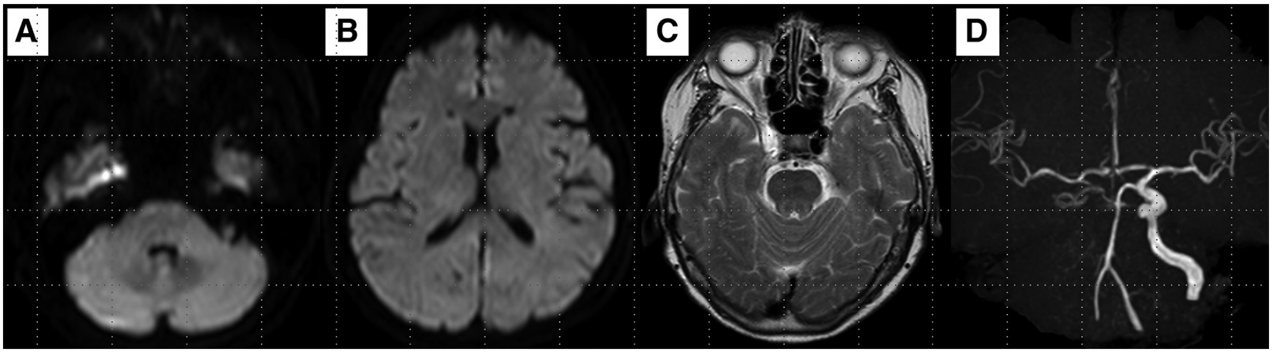

Fig. 5 MRI on the day after surgery (**A** and **B**: diffusion-weighted image) showing no high-intensity signal and 6 months after surgery (**C**: T2-weighted image and **D**: time-of-flight-MRA maximum intensity projection) showing the aneurysm had almost disappeared.

coils were tightly placed from the aneurysm-proximal area to the horizontal portion (**Fig. 4C**). After embolization, right internal carotid angiography showed no aneurysm and left internal carotid angiography demonstrated no differences in the early venous phase (**Fig. 4D**). Right vertebral arteriography showed that the territory of the right AICA (or right AICA region) was retrogradely filled by leptomeningeal anastomosis from superior cerebellar artery (SCA) and posterior inferior cerebellar artery (PICA) (**Fig. 4E**). The PTAv in this case perfused the right AICA region.

The next day, diffusion-weighted imaging was negative for ischemic complications and the FLAIR mode showed signal changes inside the aneurysm (**Fig. 5A** and **5B**). Prednisolone (30 mg/day) was started postoperatively, and although the anterior forehead pain and diplopia improved, aspirin (100 mg/day) was continued throughout the perioperative period. Six months later, MRI showed that the aneurysm had almost disappeared and diplopia was in remission (**Fig. 5C** and **5D**). The patient has had no recurrences in the 2 years after surgery.

Discussion

As far as we are aware, this is the first report of evaluating the collateral blood flow by BOT in PTAv. The details of perforating artery of PTAv are unclear, so preoperative BOT to evaluate the ischemic tolerance is important. There is no report of PTAv aneurysm recurrence by PTAv embolized incompletely. But because of a previous report featuring an ICA aneurysm in which the parent artery was occluded but the ophthalmic artery ruptured and the aneurysm recurred,[1] we chose selective embolization of the PTAv.

The PTA connects the ICA to the longitudinal neural artery during the embryonic stage,[2] and the PTAv is thought to be caused by inadequate fusion of the PTA and longitudinal neural artery.[3] However, the recent idea that PTAv stems from partial remnants of primitive lateral basilovertebral anastomosis (which temporarily connects the developing branches of the VA system)[4] has been reported and PTAv occurrence has been inconsistent, with a frequency reported to be from 0.1% to 0.3%. Nishio et al. reported that the dominant region of PTAv was anastomosis to the AICA in the majority of cases (71.6%), with anastomosis to the SCA in 28.4% and to the PICA in 18.0% of cases.[5] Weon et al. classified PTAs and PTAv into 5 types and those anastomosing to SCA, AICA, and PICA were classified as Types 5a, 5b, and 5c, respectively.[6] PTAv is also reported to be associated with cerebral aneurysms as well as PTA, with a reported incidence of about 26%.

Seven cases of cerebral aneurysm complicating PTAv have been reported to date,[5,7–12] with surgical treatment performed in two cases[8,11] and endovascular treatment in five cases.[5,7,9,10,12] Hanabusa et al. treated ruptured cerebral aneurysms associated with distal PTAv by embolizing the main trunk of the PTAv proximal to the aneurysm[7] but Yang et al. treated a spindle-shaped ruptured aneurysm associated with a proximal PTAv by embolizing the cerebral aneurysm and the PTAv proximal to the aneurysm.[12] In this case, the PTAv was perfusing the right AICA region but analysis of the right SCA and PICA revealed that the PTAv was perfusing the same region in a complementary style, leading to parent artery occlusion with no ischemic complications.

There are diverse reports that cerebral aneurysms associated with PTA are often associated with a wide neck (or other angiodysplasia) and that endovascular treatment is the first choice after a thorough preoperative examination, including BOT.[13] The evaluation points on angiography plus BOT are as follows: It is necessary to evaluate 1) whether the BA is perfused anterogradely from the VA, 2) the perfusion area of the PTAv, 3) if there is sufficient blood flow from the PcomA to the tip of the BA, and 4) if embolization with preservation of the PTAv is possible. In our case, the main trunk of the BA was not depicted by selective contrast and it was determined to be PTAv rather than PTA.

NIRS is a technique used to observe regional cerebral oxygen saturation, and Terakado et al. concluded that measurement of TOI by NIRS monitoring was useful for detecting ischemic intolerance in carotid artery stenting.[14] In our case, the use of NIRS can increase the reliability of the evaluation of possible occlusion of the ICA during BOT. However, since NIRS reflects blood flow in the frontal lobe, it is difficult to evaluate the blood flow area of PTAv.

The aneurysm in the present case had a portion of the vessels with a diameter greater than 5.5 mm, which was not an indication for the currently available flow diverter stents. Since it was also a symptomatic aneurysm, and we considered the difficulty in securing the normal ICA distal to the aneurysm. Even in the flow diverter era, parent artery occlusion certainly contributes to occlusion of aneurysms. The results of parent artery occlusion for large aneurysms are also good,[15] and it is useful as a radical treatment method for aneurysms when occlusion is possible with BOT and flow diverter is not indicated.

We chose selective embolization of the PTAv because of a previous report featuring an ICA aneurysm in which the parent artery was occluded but the ophthalmic artery ruptured and the aneurysm recurred.[1] Thus, the PTAv and the proximal ICA were embolized first but, because of the symptomatic aneurysm, we did not embolize the coils inside the aneurysm because of concerns that the compression caused by coil insertion could not be relieved. We believe that this resulted in improvement of the ophthalmoplegia and abducens nerve palsy due to the occlusion of the aneurysm.

Conclusion

ICA parent artery occlusion with PTAv selective embolization after evaluation by BOT including PTAv perfusion area is useful in the treatment of large aneurysms.

Disclosure Statement

Dr. Yuji Matsumaru has received honoraria (such as lecture fees) from Stryker, Medtronic, and Kaneka Corporation. The other authors declare no conflict of interests.

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