Cerebral Aneurysms with Internal Carotid Artery Agenesis: A Unique Case Similar to Moyamoya Disease and Literature Review

Soichiro TAKAMIYA,1,2 Tetsuyuki YOSHIMOTO,1,3 and Katsuhiko MARUICHI1

1Department of Neurosurgery, Kashiwaba Neurosurgical Hospital, Sapporo, Hokkaido, Japan
2Department of Neurosurgery, Faculty of Medicine and Graduate School of Medicine, Hokkaido University, Sapporo, Hokkaido, Japan
3Department of Neurosurgery, Hokkaido Neurosurgical Memorial Hospital, Sapporo, Hokkaido, Japan

Abstract

Internal carotid artery (ICA) agenesis/aplasia is occasionally accompanied with cerebral aneurysms caused by hemodynamic stress. If the aneurysms are located around the circle of Willis, they are managed with clipping or coil embolization. Herein, we report a case of ICA agenesis with perforating artery aneurysms treated successfully with revascularization. Moreover, a literature review of ICA agenesis with cerebral aneurysms was performed to compare with the current case. We conducted a literature review using data from PubMed. A secondary search was also performed by reviewing the references of each article previously searched. In our case, the aneurysms shrank and disappeared after direct and indirect bypass surgeries, and indirect bypass developed as in moyamoya disease (MMD). The epidemiological and clinical features of aneurysms accompanied with ICA agenesis were identified via a literature review. Aneurysms with ICA agenesis categorized as type F based on the Lie classification system, or referred to as rete mirabile, are occasionally located in an untreatable site; hence, they cannot be treated with clipping or coil embolization. Moreover, results showed that previous studies did not use revascularization for the treatment of aneurysm. In conclusion, if an aneurysm with ICA agenesis is difficult to approach directly or via an endovascular procedure, revascularization can be a treatment option.

Keywords: cerebral aneurysm, internal carotid artery agenesis, moyamoya disease, revascularization

Introduction

Internal carotid artery (ICA) agenesis or aplasia is a rare congenital anomaly, and most patients with this condition present with collateral circulations via the anterior communicating artery (Acom) and/or posterior communicating artery (Pcom).1 Patients are generally asymptomatic because of the presence of collateral circulations. However, ICA agenesis or aplasia is occasionally accompanied with cerebral aneurysms caused by hemodynamic stress. These aneurysms are found around the circle of Willis and can cause subarachnoid hemorrhage.2,3 Therefore, they are managed with direct clipping or coil embolization. In contrast, cerebral aneurysm rarely arises from a perforating artery.4 In such a case, clipping or coiling may be challenging to perform due to localization of the aneurysm.

In moyamoya disease (MMD), which is a representative disease associated with ICA occlusion and developed perforating arteries, periventricular anastomoses (PAs) sometimes develop.5 PAs are collateral vessels connecting to the periventricular medullary arteries arising from the lenticulostriate, thalamic, and choroidal arteries. The development of PAs is associated with hemorrhagic events, including intracerebral hemorrhage (ICH), in MMD.6 This relationship is similar to that between ICA agenesis with aneurysm arising from a perforating artery and aneurysm rupture. Generally, hemorrhagic MMD can be decreased by revascularization, reducing hemodynamic stress.7
Herein, we report an extremely rare case of ICA agenesis accompanied with cerebral aneurysms arising from perforating arteries. Then, direct and indirect revascularizations were performed. This study presented two interesting findings. First, the size of the aneurysms decreased after revascularization. Second, indirect bypass developed as in MMD. Moreover, a literature review of ICA agenesis with cerebral aneurysms was performed, and the current case was compared with previous cases.

**Clinical Presentation**

A 40-year-old man without significant medical and family history was brought to the emergency room due to unconsciousness. Upon arrival, his Glasgow coma scale (GCS) score was 9 (eye: 4, verbal: 1, and motor: 4). Neurological assessment showed mild left hemiparesis. Computed tomography (CT) scan revealed ICH in the right thalamus, mild high-density lesion, which was suspected as chronic ICH in the vicinity of the anterior horn of the left lateral ventricle and obstructive hydrocephalus (Figs. 1A and 1B). Moreover, it showed intraventricular hemorrhage caused by thalamic hemorrhage and obstructive hydrocephalus. Bone window CT scan at the level of the skull base revealed the absence of the right carotid canal (C, double arrows). CT: computed tomography, ICH: intracerebral hemorrhage.

Fig. 1 CT scan findings upon arrival. CT scan revealed ICH in the right thalamus (A, single arrow) and a mild high-density lesion, which was suspected as chronic ICH, in the vicinity of the anterior horn of the left lateral ventricle (B, white arrow head). Moreover, it showed intraventricular hemorrhage caused by thalamic hemorrhage and obstructive hydrocephalus. Bone window CT scan at the level of the skull base revealed the absence of the right carotid canal (C, double arrows). CT: computed tomography, ICH: intracerebral hemorrhage.

Digital subtraction angiogram (DSA) performed on the following day revealed the following (Fig. 2): (1) right ICA agenesis, (2) anastomosis from the right ascending pharyngeal artery (APhA) to the petrous potion of the ICA, (3) collateral pathway from the right accessory meningeal artery to the right anterior falcine artery via the right ophthalmic artery, (4) left anterior cerebral artery (ACA) hypoplasia, (5) collateral pathways from the M1 segment of the left middle cerebral artery (MCA) to the right ACA and MCA, (6) parenchymal anastomoses from the left MCA to the left ACA, (7) aneurysm of the parenchymal artery from the left MCA, (8) collateral pathway from the right posterior cerebral artery (PCA) to the right ICA via the right Pcom, (9) parenchymal anastomoses from the right PCA to the right MCA, and (10) a ruptured aneurysm in the right medial posterior choroidal artery. Moreover, single photon emission computed tomography (SPECT) with $^{131}$I-iodoamphetamine was performed 3 weeks after ICH, and it revealed decreased cerebral blood flow (CBF) and cerebrovascular reactivity (CVR) to acetazolamide in the right MCA and bilateral ACA territories (Supplementary Fig. 1A. All Supplementary Figures are available Online.).

These findings indicated that ICH caused the rupture of a tiny aneurysm arising from a perforating artery caused by hemodynamic stress, as in the case of hemorrhagic MMD. Considering that the aneurysms are difficult to approach with direct clipping and coil embolization and the possible pathophysiology of the aneurysms, revascularization was performed to prevent rebleeding and improve CBF. Moreover, both direct and indirect revascularizations were conducted as in MMD because the patient’s condition might be correlated with MMD. In the first revascularization, a double end-to-side anastomosis was established via the right frontotemporal craniotomy. One was located between the frontal branch of the right superficial temporal artery (STA) and the M4 segment of the right MCA, and the
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Fig. 2 DSA findings. The lateral view of the right common carotid angiography (A) revealed the tapered ICA. The lateral view of the right internal carotid angiography (B1–4) showed an abnormal anastomosis in the ICA from the APhA instead of a normal ICA. The AP view (C) and lateral view (D) of the right external carotid angiography revealed a collateral pathway from the right accessory meningeal artery to the right anterior falcine artery via the right ophthalmic artery. The AP view (E) and lateral view (F) of the left internal carotid angiography revealed the hypoplastic left ACA, collateral pathways from the M1 segment of the left MCA to the right ACA and MCA, and an aneurysm of the parenchymal artery from the left MCA (single arrow). In addition, parenchymal anastomoses from the left MCA to the left ACA were developed. AP left vertebral angiography (G: early arterial phase, H: delayed arterial phase) revealed an aneurysm in the right medial posterior choroidal artery (double arrows) and a collateral pathway from the right PCA to the right ICA via the right Pcom, which could rupture. Moreover, parenchymal anastomoses from the right PCA to the right MCA developed. ACA: anterior cerebral artery, AP: anterior–posterior, APhA: ascending pharyngeal artery, DSA: digital subtraction angiography, ICA: internal carotid artery, MCA: middle cerebral artery, PCA: posterior cerebral artery, Pcom: posterior communicating artery.
other between the temporal branch of the right STA and the M4 segment of the right MCA. The fine arteries of the brain surface were dilated and fragile, which are similar to moyamoya vessels, and the brain surface looked red. Encephalo-duro-arterio-myo-synangiosis was also performed as indirect bypass. Owing to these bypasses, the left ICA no longer needed to compensate for the right MCA territory. Hence, 2 weeks after the first revascularization, SPECT revealed mild improvement of CBF and CVR not only in the right MCA territory but also in the bilateral ACA territories, still it was not sufficient (Supplementary Fig. 1B). The second revascularization was performed 3 weeks after the first one to prevent the rupture of the aneurysm of the left MCA perforator and to improve CBF of the left ACA territory. Then, a single end-to-side anastomosis was established between the frontal branch of the left STA and the A4 segment of the left ACA via the left frontal craniotomy. The condition of the arteries in the brain surface was similar to that of the contralateral ones. Encephalo-duro-arterio-synangiosis was also performed as indirect bypass.

Postoperatively, complications, including hyper-perfusion syndrome, were not observed. SPECT revealed general improvement of CBF (Supplementary Fig. 1C). After rehabilitation, the patient was discharged without neurological deficits. After 2 years, the patient did not present with recurrent bleeding. Follow-up DSA revealed that direct anastomoses have been well established, and indirect ones have developed (Fig. 3A). In addition, the size of the aneurysm of the parenchymal artery from the left MCA decreased, and the aneurysm of the right medial posterior choroidal artery has disappeared (Figs. 3B and 3C). The patient consent for this article was provided.

Materials and Methods

Search strategy
We conducted literature review of ICA agenesis with cerebral aneurysm. The search focused on ICA agenesis, ICA aplasia, ICA hypoplasia, and carotid rete mirabile, which were categorized based on the Lie classification system.1

On April 5, 2020, a literature search on PubMed (https://www.ncbi.nlm.nih.gov/pubmed) was performed to identify case reports or case series of ICA agenesis with cerebral aneurysm. The following keywords were used: (1) “internal carotid artery” and “agenesis” and “aneurysm,” (2) “internal carotid artery” and “aplasia” and “aneurysm,” (3) “internal carotid artery” and “hypoplasia” and “aneurysm,” (4) “internal carotid artery” and “absent” and “aneurysm,” (5) “internal carotid artery” and “absence” and “aneurysm,” and (6) “rete” and “humans”[mesh] and “aneurysm.”

The search was restricted to articles written in English or Japanese. We included cases from 1957 to 2019. The articles about MMD, acquired ICA occlusion, and ICA agenesis with primitive carotid-basilar anastomosis as the main collateral pathway were excluded. After deleting duplicates, the abstracts of articles were screened to exclude those not meeting the criteria. Then, full-text screening was performed. A secondary search was carried out by reviewing the references of each article searched previously. The abovementioned inclusion criteria were applied. The search strategy and selection process of the articles are shown in Supplementary Fig. 2.

Results
In total, 85 articles, including 95 cases of ICA agenesis with cerebral aneurysm, were included in the analysis. Data on age, sex, laterality of the lesion, Lie’s classification, onset, aneurysmal site, and treatment for aneurysm were reviewed. Whether the aneurysmal site was ipsilateral and contralateral to the ICA agenesis was identified. For convenience, we considered aneurysms with bilateral ICA agenesis as contralateral. Data are shown in Table 1.2,4,8–90

The mean age of the patients was 42.5 (range: 0–81) years. Of 95 patients, 35 (36.8%) were men and 56 (58.9%) women. However, the sex of four patients was not identified. The proportion of female patients was 1.6 times higher than that of male patients (Supplementary Fig. 3A). In terms of the laterality of the lesions, 28 (29.5%) patients presented with right-side lesions, 47 (49.5%) with left-side lesions, and 20 (21.1%) with lesions in both sides (Supplementary Fig. 3B). Based on the Lie classification system, 38 (40.0%) lesions were categorized as type A, 16 (16.8%) as type B, 14 (14.7%) as type C, 12 (12.6%) as type D, and 13 (13.7%) as type F. However, the lesions in two patients were not classified (Supplementary Fig. 3C). The most common manifestation of ICA agenesis with aneurysm was subarachnoid hemorrhage (66.3%), followed by headache without intracranial hemorrhage (8.4%) and ischemic symptoms (including cerebral infarction, transient ischemic attack, and amaurosis fugax) (7.4%) (Supplementary Fig. 3D).

In term of the number of aneurysms, 76 (80.0%) patients presented with a single aneurysm, and 19 (20.0%) with multiple ones. Of 113 aneurysms (counted separately as multiple aneurysms), Acom aneurysms were the most common (38.9%, including one case involving the azygos ACA), followed by

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basilar artery (21.2%) and contralateral ICA (12.4%) (Supplementary Fig. 3E). Moreover, the frequent aneurysmal site of each lesion classified using the Lie classification system was assessed (Fig. 4). Results showed relationships between Lie’s classification and aneurysmal site, which were as follows: (1) more than 50% of aneurysms were found in the Acom in types A, B, and D; (2) contralateral ICA aneurysms were also common in type B; (3) ipsilateral MCA aneurysms were observed only in types A and D. However, contralateral MCA aneurysms were not detected; (4) all aneurysms in type C were noted in the posterior circulation; and (5) there was no predisposition for an aneurysmal site in type F. The most common treatment for aneurysms was aneurysmal clipping (47.7%). In total, 26 (27.4%) patients, including those with extremely severe condition, received conservative therapies prior to receiving invasive treatment. In total, 13 (13.7%) patients underwent coil embolization (Supplementary Fig. 3F). Revascularization was not performed for the treatment of ICA agenesis with cerebral aneurysm.

**Discussion**

ICA agenesis occurs at an early stage during embryonic development, and several types of collateral pathways develop to compensate for CBF. This condition is classified according to collateral circulations. The Lie classification system, which is the most popular classification system, describes six types of collateral pathways, which are as follows:

Type A, unilateral absence of the ICA with a collateral pathway to the ipsilateral ACA via the Acom and the ipsilateral MCA via the Pcom; type B, unilateral absence of the ICA with a collateral pathway to the ipsilateral ACA and MCA via the Acom; type C, bilateral absence of ICAs with a collateral pathway to the ACAs and MCAs via the Pcoms; type D, unilateral absence of the cervical portion of the ICA with the intercavernous collateral pathway from the cavernous portion of the contralateral ICA; type E, ACAs supplied by the bilateral hypoplastic ICAs and MCAs supplied via the Pcoms; and type F, transcranial anastomoses from the
### Table 1 The summary of ICA aplasia with aneurysms

| Author, year | Age, sex | Agenesis/aplasia side | Onset | Lie type | Aneurysm localization | Treatment |
|--------------|----------|-----------------------|-------|----------|-----------------------|-----------|
| Kwak, 2019   | 57, F    | Left                  | IS    | A        | BA, right ACA          | Coil      |
| Agarwal, 2019| 67, M    | Bilateral             | H     | C        | BA, left PCA-Pcom      | ND        |
| Hou, 2019    | 58, M    | Left                  | IS    | D        | BA                    | Conservation |
| Chen, 2019   | 39, F    | Bilateral             | SAH   | F        | Left PCA collateral    | Conservation (moyamoya like) |
| Shukla, 2018 | 60, M    | Left                  | H     | A        | Acom                  | Conservation |
| Verma, 2018  | 28, F    | Right                 | H     | F        | Right ICA, left PCA    | ND        |
| Kumagai, 2017| 47, M    | Left                  | others | D       | Acom (multiple)        | Clip      |
| Ohtani, 2017 | 68, F    | Bilateral             | H     | C        | BA-left SCA, right PCA-Pcom | Conservation |
| Dinca, 2017  | 62, F    | Right                 | SAH   | B        | Left ICA-Ophth.        | Clip      |
| Cruz, 2017   | 74, ND   | Bilateral             | SAH   | C        | BA                    | Clip      |
| Alurkar, 2016| 39, F    | Right                 | SAH   | D        | Right MCA             | Clip      |
| Mohan, 2015  | 49, F    | Right                 | SAH   | B        | Left ICA-Ophth.        | Clip      |
| Yamasaki, 2015| 50, M   | Left                  | SAH   | B        | Right ICA-Acho.        | Clip      |
| Paschoal, 2015| 28, F   | Right                 | SAH   | F        | Left ICA              | Clip      |
| Cherungottil, 2014| 7, F | Right                 | NP    | A        | Right PCA-Pcom (thrombosed) | Conservation |
| Nagahata, 2013| 70, M   | Bilateral             | SAH   | F        | Bilateral RMA          | Coil      |
| Kang, 2012   | 59, F    | Left                  | SAH   | A        | ACA (infraoptic azygous portion) | Coil |
| Kim, 2012    | 43, M    | Bilateral             | SAH   | F        | BA                    | Clip      |
| Lim, 2012    | 41, F    | Left                  | SAH   | A        | BA                    | Clip      |
| Bhaskar, 2012| 32, M    | Left                  | SAH   | A        | Acom                  | Clip      |
| Siddiqui, 2012| 48, F  | Bilateral             | SAH   | C        | Right PCA-Pcom        | Conservation |
| Pasaoglu, 2011| 61, M  | Left                  | IS    | A        | Left VA-BA (multiple)  | Conservation |
| Wani, 2011   | 50, M    | Left                  | SAH   | A        | Right ICA (paraclinoid), Acom | Clip |
| Akiyama, 2010| 63, F    | Left                  | H     | A        | Left MCA, BA          | Clip, coil |
| Xie, 2010    | 45, M    | Left                  | others | B       | Right ICA top         | Clip      |
| Elazab, 2010 | 0, F     | Right                 | SAH   | A        | BA                    | Conservation |
| Barbosa, 2010| 34, F    | Bilateral             | SAH   | C        | BA                    | Coil      |
| Kim, 2009    | 56, F    | Bilateral             | SAH   | C        | Right PICA            | Clip      |
| Erdem, 2009  | 49, M    | Bilateral             | SAH   | C        | Right PCA, BA-left SCA| Clip      |
| Suyama, 2009 | 69, F    | Left                  | SAH   | D        | Acom                  | Clip      |
| Chen, 2008   | 53, F    | Right                 | SAH   | B        | Acom                  | Conservation |
| Kazumata, 2008| 39, F   | Bilateral             | SAH   | C        | Left PCA-Pcom          | Clip      |
| Horie, 2008  | 55, F    | Left                  | Others | D       | Acom                  | Clip      |
| Demirgil, 2007| 18, F  | Left                  | SAH   | A        | Acom                  | Clip      |
| Orakdogen, 2007| 43, F | Left                  | SAH   | A        | Acom                  | Clip      |
| Zink, 2007   | 49, M    | Right                 | Other  | D        | Right MCA, Acom       | Clip      |
| Henkes, 2007 | 36, M    | Left                  | SAH   | F        | Left ICA, left PCA    | Clip      |
| Henkes, 2007 | 54, M    | Right                 | SAH   | F        | Acom                  | Clip      |
Table 1 The summary of ICA aplasia with aneurysms (Continued)

| Author, year | Age, sex | Agenesis/ aplanasia side | Onset | Lie type | Aneurysm localization | Treatment |
|-------------|----------|-------------------------|-------|----------|----------------------|-----------|
| Wong, 43    | 81, F    | Left                    | SAH   | A        | Acom                 | Conservation |
| Funii, 44   | 56, M    | Right                   | Others| A        | Acom                 | Clip       |
| Herwadkar, 65 | 52, F | Bilateral              | SAH   | F        | BA                   | Coil       |
| Briganti, 46 | 56, F  | Bilateral              | Others| C        | BA                   | Conservation |
| Gailloud, 47 | 53, F  | Right                   | IS    | B        | Left ACA             | Clip       |
| Amano, 48   | 42, F    | Left                    | SAH   | A        | Right ICA            | Clip       |
| Tasar, 49   | 17, M    | Left                    | SAH   | A        | Left MCA             | ND         |
| Bodhey, 50  | 57, M    | Right                   | IS    | D        | Right PCA-Pcom       | Clip       |
| Lee, 51     | 55, M    | Right                   | SAH   | A        | Acom                 | Clip       |
|             | 19, F    | Left                    | SAH   | A        | Acom                 | Clip       |
|             | 51, M    | Right                   | SAH   | A        | Acom                 | Clip       |
|             | 57, F    | Left                    | H     | A        | Acom                 | Clip       |
|             | 50, F    | Right                   | IS    | B        | Left ICA, right PCA  | Conservation |
|             | 31, M    | Bilateral              | H     | C        | BA                   | Coil       |
| Okita, 52   | 44, F    | Bilateral              | SAH   | C        | BA-right AICA        | Clip       |
| Ide, 53     | 74, F    | Left                    | Others| B        | Right ICA (paracalinoi) | Conservation |
| Jordan, 54  | 10, M    | Left                    | Others| D        | Right ICA            | Conservation |
| Florio, 55  | 62, F    | Left                    | SAH   | A        | Acom                 | Clip       |
| Czarnecki, 56 | 45, M  | Left                    | Others| B        | Acom (multiple)      | Clip       |
| Sugiu, 57   | 65, F    | Right                   | SAH   | A        | BA                   | Conservation |
|             | 67, F    | Right                   | Others| A        | BA                   | Coil       |
| Tanaka, 58  | 45, M    | Left                    | SAH   | F        | Acom                 | Clip       |
| Armand, 59  | ND       | Left                    | SAH   | B        | Acom                 | ND         |
| Ide, 60     | 38, F    | Right                   | SAH   | A        | Right PCA-Pcom       | Clip       |
| Ito, 61     | 42, F    | Left                    | SAH   | F        | Right PCA-Pcom, left ICA | Clip |
| Rondepierre, 62 | 34, F | Right                   | IS    | F        | Right ICA            | Conservation |
| Nakai, 63   | 27, M    | Right                   | SAH   | B        | Acom                 | Clip       |
| Quint, 64   | 60, F    | Right                   | NP    | D        | Left ICA, right MCA, Acom | Conservation |
| Yoshida, 65 | 67, F    | Left                    | SAH   | B        | Acom                 | Clip       |
| Anegawa, 66 | 52, F    | Bilateral              | H     | C        | Left PCA             | Clip       |
| Afifi, 67   | 0, F     | Left                    | others| B        | Acom                 | Clip       |
| Kunishiro, 68 | 70, M | Left                    | SAH   | A        | Acom, left MCA, BA   | Clip       |
| Petreia, 69 | 53, F    | Right                   | SAH   | A        | Acom                 | Clip       |
| Tracy, 70   | 34, M    | Right                   | SAH   | D        | Acom                 | Clip       |
| Amacher, 71 | 15, M    | Bilateral              | ND    | C        | BA                   | Clip       |
| Huber, 72   | 26, ND   | Left                    | SAH   | D        | Acom                 | Clip       |
| Bernini, 73 | 38, F    | Right                   | SAH   | A        | Acom                 | Conservation |
| Shigemori, 74 | 48, F  | Left                    | SAH   | A        | Acom                 | Clip       |
| Katakura, 75 | 41, F  | Left                    | Others| ND       | BA                   | Conservation |
| Waga, 76    | 60, F    | Left                    | SAH   | A        | Acom                 | Clip       |
external carotid artery to the ICA, which is referred to as rete mirabile. This condition is commonly observed in lower mammals, such as pigs, cows, sheep, and goats. However, it is not typical among humans. Araki et al. described the angiographic features of rete mirabile, which can be used for diagnosis, and the characteristics were as follows: (1) hypoplasia of the ICA, (2) arterial anastomosis between the internal maxillary artery (IMA) and the cavernous portion of the ICA, (3) dilation of the ophthalmic artery, (4) supraclinoid ICA supplied by the IMA and ophthalmic artery, (5) bilateral lesions, and (6) absence of intradural anastomoses as in MMD. In the current case, the patient did not meet these criteria because the lesion was unilateral and the arterial anastomosis was located between the APhA and the petrous potion of the ICA. Therefore, it was difficult to diagnose the present case as rete mirabile. Meanwhile, we considered our case as type F based on the Lie classification because he had an anastomosis from the APhA to the ICA.

The incidence of cerebral aneurysms is high in ICA agenesis. Meanwhile, the normal incidence rate of cerebral aneurysms is only 2%–4%, and that in patients with ICA agenesis is 25%–67%, 2.51,68,74,79,81 It is caused by hemodynamic stress in the collateral circulation pathways. Our review has revealed the common aneurysmal sites for each Lie’s classification, which nearly always exist on the collateral pathway to compensate for the territory that should be perfused by the aplastic ICA. It will support abovementioned information that hemodynamic burden causes the high incidence of aneurysm accompanied with ICA agenesis.

In our review, the radical treatments for aneurysm were direct clipping or coil embolization. These treatments could be performed because the aneurysms were commonly found around the circle of Willis. By contrast, an aneurysm with rete mirabile is occasionally located in a unique site, which is difficult to approach, as in the current case and in some cases. Even if an aneurysm can be approached, the current artery cannot be easily saved by direct clipping or coil embolization. Therefore, these treatments are not used. In the current case, the aneurysms disappeared after revascularizations. Similar results were observed in aneurysms with MMD or ICA occlusion (including those of unknown etiology). Moreover, revascularization was also effective for aneurysms with moyamoya syndrome associated with cranial irradiation. Bypass surgery could be an effective treatment for an aneurysm as it reduces hemodynamic stress. Indeed, there are some reports that the aneurysms located in collateral vessels could

| Author, year | Age, sex | Agenesis/aplasia side | Onset | Lie type | Aneurysm localization | Treatment |
|-------------|----------|----------------------|-------|----------|-----------------------|-----------|
| Tsuruta, 1977 | 48, M | Left | SAH | A | Acom | Clip |
| Naito, 1977 | 12, F | Left | SAH | A | Acom | Clip |
| Servo, 1977 | 48, M | Left | SAH | A | Right ICA | Conservation |
| Rosen, 1975 | 37, M | Bilateral | SAH | C | BA | Conservation |
| Teal, 1973 | 66, F | Left | Others | A | Right ICA, Acom | Evac. Hx |
| Sakurai, 1972 | 66, M | Right | SAH | A | Acom | EVD |
| Handa, 1971 | 28, F | Left | SAH | B | Right ICA-Ophth. | Conservation |
| Tangchaid, 1970 | 30, F | Left | SAH | A | Left MCA | Conservation |
| Moyes, 1969 | 37, F | Left | SAH | A | BA | Clip |
| Lhermitte, 1968 | 63, F | Left | SAH | A | Acom | Conservation |
| Hawkins, 1967 | 37, M | Bilateral | SAH | F | BA | Conservation |
| Burnester, 1961 | 43, M | Right | SAH | A | Acom | Muscle wrap |
| Lagarde, 1957 | 42, M | Right | SAH | B | Acom | Muscle wrap |
| Cohen, 1957 | ND | Left | ND | ND | Acom | ND |

AICA: anterior inferior cerebellar artery, ACA: anterior cerebral artery, Acho: anterior choroidal artery, Acom: anterior communicating artery, BA: basilar artery, evac. Hx: evacuation of a hematoma, F: female, H: headache without subarachnoid hemorrhage, ICA: internal carotid artery, IS: ischemic stroke (including cerebral infarction, transient ischemic attack, and amaurosis fugax), M: male, MCA: middle cerebral artery, NC: nerve palsy, ND: not described, ophth: ophthalmic artery, PCA: posterior cerebral artery, Pcom: posterior communicating artery, PICA: posterior inferior cerebellar artery, RMA: radiculomedullary artery, SAH: subarachnoid hemorrhage, SCA: superior cerebellar artery, VA: vertebral artery.
be disappeared by conservative therapy.\textsuperscript{102,103} However, Kanamori et al.\textsuperscript{97} reported the importance of early surgical revascularization to prevent re-rupture of collateral artery aneurysms. Hence, we selected revascularization rather than conservative therapy for the present case. In addition, the aneurysmal sites in the current case were similar to those in PAs in MMD, which are associated with hemorrhagic event in MMD.\textsuperscript{6} The Japan Adult Moyamoya (JAM) trial showed that revascularization can decrease the incidence of hemorrhagic MMD. In addition, a supplementary analysis of the JAM trial also showed revascularization for MMD with hemodynamic failure could significantly prevent the hemorrhagic event and proposed that hemodynamic failure should be considered for the surgical indication in hemorrhagic MMD.\textsuperscript{104} Therefore, selective revascularization is considered reasonable based on the cerebrovascular assessment results in our case. We believe that hemodynamic failure was an important finding to predict a postoperative result and decide a surgical indication. However, whether the aneurysms were true aneurysms or pseudoaneurysms could not be validated, and this is considered a limitation of the study.

Only one report regarding ICA agenesis with aneurysm discussed about CBF.\textsuperscript{24} Therefore, the proportion of patients whose CBF decreased was not identified. Considering the report of Sugiura et al.,\textsuperscript{57} which was described about de novo aneurysm associated with agenesis of ICA, it is possible that some patients who undergo direct clipping or coil embolization for cerebral aneurysm are at risk of recurrence or de novo aneurysm caused by hemodynamic stress. Thus, these patients must be follow-up regularly. In addition, performing a cerebral perfusion examination should be considered, and revascularization must be performed if appropriate.

**Conclusions**

ICA agenesis is occasionally accompanied with cerebral aneurysms. Most aneurysms are found adjacent to the circle of Willis. However, they rarely arise apart from this structure, particularly in cases involving lesions classified as type F, also known as carotid rete mirabile. Aneurysms are difficult to approach directly or via an endovascular procedure in such cases. Therefore, revascularization can be a treatment option, as in the current case.

Whether de novo aneurysms occur after surgery should be monitored not only in cases in which aneurysms are difficult to approach but also in cases in which aneurysms are easy to approach. In some cases, cerebral perfusions should be assessed because cerebral misery perfusions, which can cause both ischemic and hemorrhagic strokes, may occur.
Conflicts of Interest Disclosure

The authors report no conflict of interest associated with this paper. All authors have registered online self-reported COI Disclosure Statement Forms through the website for JNS members.

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Corresponding author: Soichiro Takamiya, MD
Department of Neurosurgery, Faculty of Medicine and Graduate School of Medicine, Hokkaido University, North 15 West 7, Kita-ku, Sapporo, Hokkaido 060-8638, Japan.
e-mail: soichiro.tkmy@gmail.com