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

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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]

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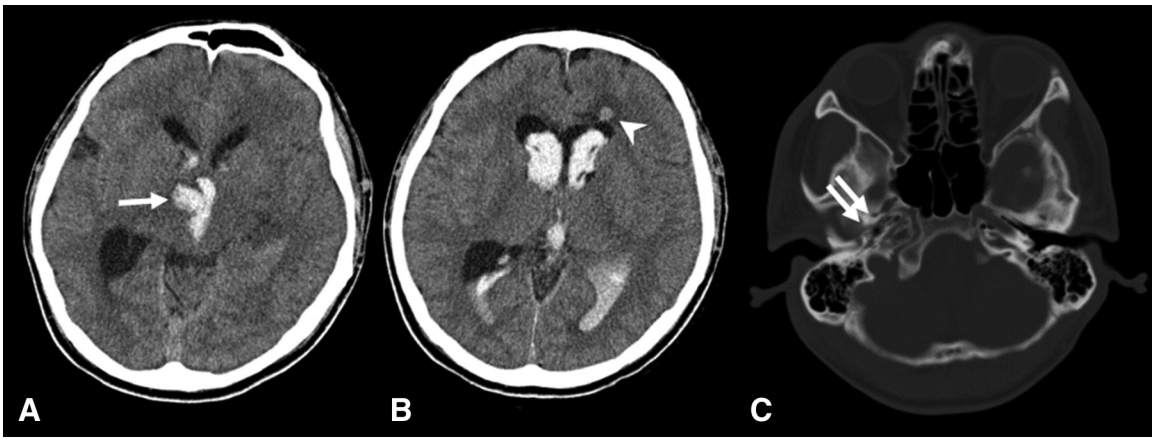


Fig. 1 CT scan findings upon arrival. CT scan revealed ICH in the right thalamus (A, single arrow) and a mild high-density lesion, which was suspected as chronic ICH, in the vicinity of the anterior horn of the left lateral ventricle (B, white arrow head). Moreover, it showed intraventricular hemorrhage 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.

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, intraventricular hemorrhage, and obstructive hydrocephalus (Figs. 1A and 1B). In addition, the right carotid canal was not detected (Fig. 1C). Emergent external ventricular drainage was performed, and the patient's GCS score improved immediately from 9 to 14.

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 ^{123}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

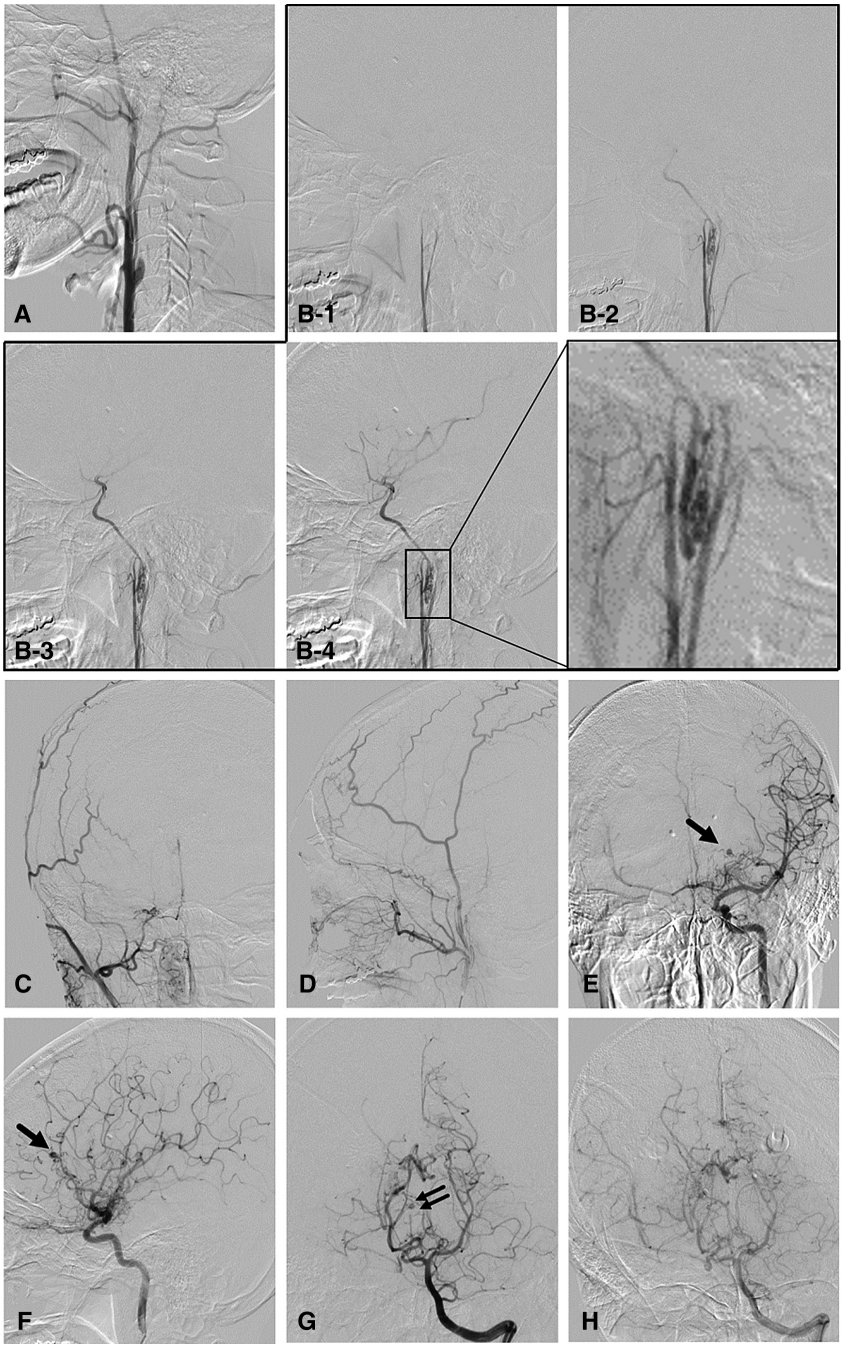


Fig. 2 DSA findings. The lateral view of the right common carotid angiography (A) revealed the tapered ICA. The lateral view of the right internal carotid angiography (B1–4) showed an abnormal anastomosis in the ICA from the APhA instead of a normal ICA. The AP view (C) and lateral view (D) of the right 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 hyperperfusion 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

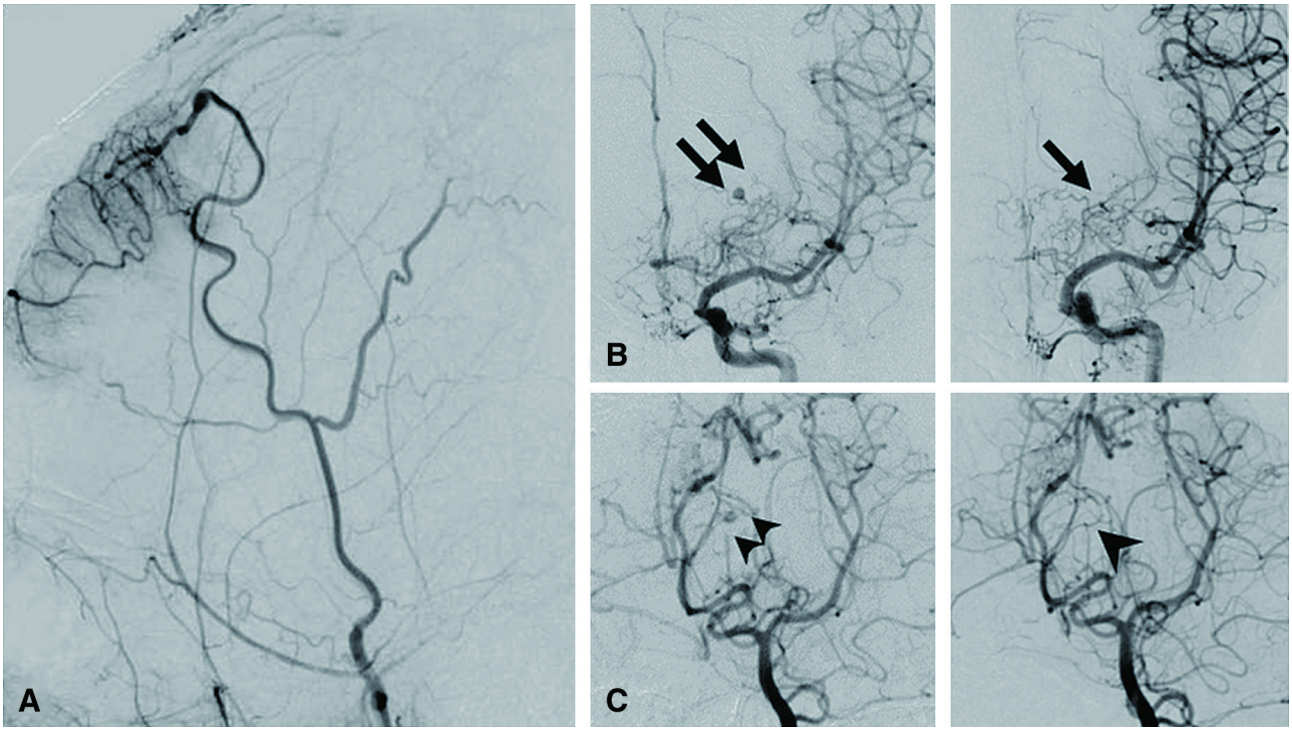


Fig. 3 DSA findings 7 months after surgery. The lateral view of the left external carotid angiography revealed the development of collateral arteries from the frontal branch of the middle meningeal artery which was the donor of the bypass (A). The AP view of the left internal carotid angiography revealed that the size of the aneurysm of the parenchymal artery from the left MCA decreased (B. right, single arrow) compared with preoperative DSA (B. left, double arrows). The AP view of the left vertebral angiography revealed that the aneurysm in the right medial posterior choroidal artery disappeared after surgery (C. right, single arrowhead) compared with image before the surgery (C. left, double arrow heads). AP: anterior–posterior, DSA: digital subtraction angiography, MCA: middle cerebral artery.

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 Pcom; 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,[8] 2019	57, F	Left	IS	A	BA, right ACA	Coil
Agarwal,[9] 2019	67, M	Bilateral	H	C	BA, left PCA-Pcom	ND
Hou,[10] 2019	58, M	Left	IS	D	BA	Conservation
Chen,[4] 2019	39, F	Bilateral	SAH	F	Left PCA collateral (moyamoya like)	Conservation
Shukla,[11] 2018	60, M	Left	H	A	Acom	Conservation
Verma,[12] 2018	28, F	Right	H	F	Right ICA, left PCA	ND
	34, F	Bilateral	SAH	F	BA, right PCA	Clip
Kumagai,[13] 2017	47, M	Left	others	D	Acom (multiple)	Clip
Ohtani,[14] 2017	68, F	Bilateral	H	C	BA-left SCA, right PCA-Pcom	Conservation
Dinca,[15] 2017	62, F	Right	SAH	B	Left ICA-Ophth.	Clip
Cruz,[16] 2017	74, ND	Bilateral	SAH	C	BA	Clip
Alurkar,[17] 2016	39, F	Right	SAH	D	Right MCA	Clip
Mohan,[18] 2015	49, F	Right	SAH	B	Left ICA-Ophth.	Clip
Yamasaki,[19] 2015	50, M	Left	SAH	B	Right ICA-Acho.	Clip
Paschoal,[20] 2015	28, F	Right	SAH	F	Left ICA	Clip
Cherungottil,[21] 2014	7, F	Right	NP	A	Right PCA-Pcom (thrombosed)	Conservation
Nagahata,[22] 2013	70, M	Bilateral	SAH	F	Bilateral RMA	Coil
Kang,[23] 2012	59, F	Left	SAH	A	ACA (infraoptic azygous portion)	Coil
Kim,[24] 2012	43, M	Bilateral	SAH	F	BA	Coil
Lim,[25] 2012	41, F	Left	SAH	A	BA	Coil
Bhaskar,[26] 2012	32, M	Left	SAH	A	Acom	Clip
Siddiqui,[27] 2012	48, F	Bilateral	SAH	C	Right PCA-Pcom	Conservation
Pasaoglu,[28] 2011	61, M	Left	IS	A	Left VA~BA (multiple)	Conservation
Wani,[29] 2011	50, M	Left	SAH	A	Right ICA (paraclinoid), Acom	Clip
Akiyama,[30] 2010	63, F	Left	H	A	Left MCA, BA	Clip, coil
Xie,[31] 2010	45, M	Left	others	B	Right ICA top	Clip
Elazab,[32] 2010	0, F	Right	SAH	A	BA	Conservation
Barbosa,[33] 2010	34, F	Bilateral	SAH	C	BA	Coil
Kim,[34] 2009	56, F	Bilateral	SAH	C	Right PICA	Clip
Erdem,[35] 2009	49, M	Bilateral	SAH	C	Right PCA, BA-left SCA	Clip
Suyama,[36] 2009	69, F	Left	SAH	D	Acom	Clip
Chen,[37] 2008	53, F	Right	SAH	B	Acom	Conservation
Kazumata,[38] 2008	39, F	Bilateral	SAH	C	Left PCA-Pcom	Clip
Horie,[39] 2008	55, F	Left	Others	D	Acom	Clip
Demirgil,[40] 2007	18, F	Left	SAH	A	Acom	Clip
Orakdogen,[41] 2007	43, F	Left	SAH	A	Acom	Clip
Zink,[2] 2007	49, M	Right	Other	D	Right MCA, Acom	Coil
Henkes,[42] 2007	36, M	Left	SAH	F	Left ICA, left PCA	Coil
	54, M	Right	SAH	F	Acom	Coil

Table 1 The summary of ICA aplasia with aneurysms (Continued)

Author, year	Age, sex	Agenesis/ aplasia side	Onset	Lie type	Aneurysm localization	Treatment
Wong,[43] 2006	81, F	Left	SAH	A	Acom	Conservation
Funiu,[44] 2006	56, M	Right	Others	A	Acom	Clip
Herwadkar,[45] 2006	52, F	Bilateral	SAH	F	BA	Coil
Briganti,[46] 2004	56, F	Bilateral	Others	C	BA	Conservation
Gailloud,[47] 2004	53, F	Right	IS	B	Left ACA	Clip
Amano,[48] 2004	42, F	Left	SAH	A	Right ICA	Clip
Taşar,[49] 2004	17, M	Left	SAH	A	Left MCA	ND
Bodhey,[50] 2004	57, M	Right	IS	D	Right PCA-Pcom	Clip
Lee,[51] 2003	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] 2001	44, F	Bilateral	SAH	C	BA-right AICA	Coil
Ide,[53] 2000	74, F	Left	Others	B	Right ICA (paraclinoid)	Conservation
Jordan,[54] 2000	10, M	Left	Others	D	Right ICA	Conservation
Florio,[55] 1999	62, F	Left	SAH	A	Acom	Clip
Czarnecki,[56] 1998	45,M	Left	Others	B	Acom (multiple)	Clip
Sugiura,[57] 1997	65, F	Right	SAH	A	BA	Conservation
	67, F	Right	Others	A	BA	Coil
Tanaka,[58] 1996	45, M	Left	SAH	F	Acom	Clip
Armand,[59] 1996	ND	Left	SAH	B	Acom	ND
Ide,[60] 1995	38, F	Right	SAH	A	Right PCA-Pcom	Clip
Ito,[61] 1994	42, F	Left	SAH	F	Right PCA-Pcom, left ICA	Clip
Rondepierre,[62] 1993	34, F	Right	IS	F	Right ICA	Conservation
Nakai,[63] 1992	27, M	Right	SAH	B	Acom	Clip
Quint,[64] 1989	60, F	Right	NP	D	Left ICA, right MCA, Acom	Conservation
	65, F	Left	SAH	D	Acom	Clip
Yoshida,[65] 1988	67, F	Left	SAH	B	Acom	Conservation
Anegawa,[66] 1987	52, F	Bilateral	H	C	Left PCA	Clip
Afifi,[67] 1987	0, F	Left	others	B	Acom	Clip
Kunishiro,[68] 1987	70, M	Left	SAH	A	Acom, left MCA, BA	Clip
Petrela,[69] 1987	53, F	Right	SAH	A	Acom	Clip
Tracy,[70] 1987	34, M	Right	SAH	D	Acom	Clip
Amacher,[71] 1981	15, M	Bilateral	ND	C	BA	Clip
Huber,[72] 1980	26, ND	Left	SAH	D	Acom	Clip
Bernini,[73] 1980	38, F	Right	SAH	A	Acom	Conservation
Shigemori,[74] 1980	48, F	Left	SAH	A	Acom	Clip
Katakura,[75] 1979	41, F	Left	Others	ND	BA	Conservation
Waga,[76] 1978	60, F	Left	SAH	A	Acom	Clip

Table 1 The summary of ICA aplasia with aneurysms (Continued)

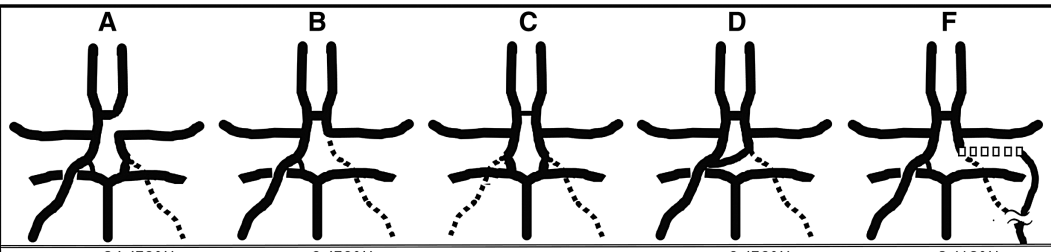
Author, year	Age, sex	Agenesis/ aplasia side	Onset	Lie type	Aneurysm localization	Treatment
Tsuruta,[77] 1977	48, M	Left	SAH	A	Acom	Clip
Naito,[78] 1977	12, F	Left	SAH	A	Acom	Clip
Servo,[79] 1977	48, M	Left	SAH	A	Right ICA	Conservation
Rosen,[80] 1975	37, M	Bilateral	SAH	C	BA	Conservation
Teal,[81] 1973	66, F	Left	Others	A	Right ICA, Acom	Evac. Hx
Sakurai,[82] 1972	66, M	Right	SAH	A	Acom	EVD
Handa,[83] 1971	28, F	Left	SAH	B	Right ICA-Ophth.	Conservation
Tangchai,[84] 1970	30, F	Left	SAH	A	Left MCA	Conservation
Moyes,[85] 1969	37, F	Left	SAH	A	BA	Clip
Lhermitte,[86] 1968	63, F	Left	SAH	A	Acom	Conservation
Hawkins,[87] 1967	37, M	Bilateral	SAH	F	BA	Conservation
Burmester,[88] 1961	43, M	Right	SAH	A	Acom	Muscle wrap
	32, F	Left	SAH	B	Acom	Muscle wrap
Lagarde,[89] 1957	42, M	Right	SAH	B	Acom	Muscle wrap
Cohen,[90] 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.

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.[91] However, it is not typical among humans. Araki et al.[92] 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.[4,12] 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[93–97] or ICA occlusion (including those of unknown etiology).[98–100] Moreover, revascularization was also effective for aneurysms with moyamoya syndrome associated with cranial irradiation.[101] 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



Lie's classification		A	B	C	D	F
ACA	Acom	24 (53%)	9 (50%)		9 (56%)	2 (12%)
	Ipsi					
	Contra	1 (2%)	1 (6%)			
ICA	Ipsi					4 (24%)
	Contra	4 (9%)	7 (39%)		2 (13%)	1 (6%)
MCA	Ipsi	4 (9%)			3 (19%)	
	Contra					
BA		9 (20%)		10 (59%)	1 (6%)	4 (24%)
PCA	Ipsi	2 (4%)	1 (6%)	6 (35%)	1 (6%)	2 (12%)
	Contra					2 (12%)
Others		1 (2%)		1 (6%)		2 (12%)

Fig. 4 Common aneurysmal site for each Lie's classification. In Lie's type A aneurysms, 53% arose from the Acom (including one case involving the azygos ACA) and 20% from the BA. In Lie's type B aneurysms, 50% arose from the Acom and 39% from the contralateral ICA, which had a unique characteristic compared with the other types. In Lie's type C aneurysms, 59% arose from the BA and 35% from the ipsilateral PCA. All aneurysms of this type existed posterior circulation. In Lie's type D aneurysms, 56% arose from the Acom. In Lie's type F aneurysms, there was no predisposition for an aneurysmal site. ACA: anterior cerebral artery, Acom: anterior communicating artery, BA: basilar artery, ICA: internal carotid artery, PCA: posterior cerebral artery.

be disappeared by conservative therapy.[102,103] However, Kanamori et al.[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.[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.[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.[24] Therefore, the proportion of patients whose CBF decreased was not identified. Considering the report of Sugiura et al.,[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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