Endovascular Embolization of Distal Anterior Choroidal Artery Aneurysms Associated with Moyamoya Disease

A Report of Two Cases and a Literature Review

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Summary

We evaluated the feasibility of endovascular embolization for the management of distal anterior choroidal artery (AChA) aneurysms associated with moyamoya disease and performed a literature review to summarize their clinical features and treatment. We describe two cases of moyamoya disease-associated distal AChA aneurysms treated by endovascular embolization. In both cases, a good outcome was observed. We performed a MEDLINE (1980–2010) search which identified 13 similar cases. Our analysis of the clinical data from these 15 cases led us to conclude that (i) endovascular embolization is an effective and feasible treatment for distal AChA aneurysms associated with moyamoya disease; (ii) aneurysm location and the preservation of the parent artery are two major prognostic factors for moyamoya disease-associated distal AChA aneurysms subjected to craniotomy or endovascular therapy; (iii) the parent artery should be preserved when the aneurysm is located in the temporal horn of the lateral ventricle, but sacrificed when it is located in the trigone of the lateral ventricle.

Introduction

Moyamoya disease is a rare disorder characterized by blockage or occlusion of one or, usually, both internal carotid arteries. When the anterior choroidal artery (AChA) is not involved in moyamoya disease, the trunk and

branches of the AChA often develop compensatory dilation and extend to distal regions, thereby forming collateral vessels that increase blood flow. However, the increase in blood flow greatly increases the probability of the occurrence of distal AChA aneurysms¹. Distal AChA aneurysms are quite rare, often associated with moyamoya disease and there is little knowledge concerning their treatment².

Between 2000 and 2010, we encountered two cases of distal AChA aneurysms associated with moyamoya disease. Both of these patients underwent endovascular embolization and showed a good clinical outcome. Since distal AChA aneurysms are quite rare, we performed a literature review to summarize the clinical features and treatment of AChA aneurysms. Our MEDLINE search (1980–2010) yielded another 13 cases of moyamoya disease-associated distal AChA aneurysms^{3–12}, including five cases in which the patients underwent craniotomy and 1 case in which the patient underwent endovascular treatment.

Although craniotomy is considered to be a traditional treatment for distal AChA aneurysms associated with moyamoya disease, it can gravely damage nervous tissue. Furthermore, these aneurysms are difficult to identify and expose during craniotomy¹³.

In recent years, attention has turned to the potential of endovascular techniques in the treatment of aneurysms. Digital subtraction angiography (DSA)-guided endovascular intervention has the advantages of minimal invasiveness and accurate localization, and has been

Figure 1 Case 1. A) CT scan revealing left intraventricular hemorrhage involving the third ventricle and the contralateral lateral ventricle. B, C) DSA images showing moyamoya vessels. D, E) DSA and 3D-DSA images showing an aneurysm (arrow) on the left distal AChA. F) DSA image showing compensatory collateral circulation and increased blood-supply area. G) DSA image revealing the angiographic features of the AChA. H) DSA image showing aneurysm disappearance after Glubran 2 embolization. I) DSA image at follow-up showing the development of distal collateral circulation (ellipse) and no evidence of aneurysm recanalization.

widely applied in clinical practice since it was first reported in 1991^{14,15}.

Currently, little is known as to whether different treatments (craniotomy and endovascular therapy) differ with respect to outcome in

terms of the prognosis of distal AChA aneurysms associated with moyamoya disease. In order to explore the feasibility of endovascular embolization in the management of such aneurysms, here we report two cases of moyamoya

Figure 2 Case 2. A) CTA image revealing a cystic aneurysm in the distal AChA. B, C) DSA images showing intracranial moyamoya vessels. D) DSA image revealing an aneurysm (arrow) on the left distal AChA. E) DSA image showing the development of collateral circulation and no aneurysm. F, G) DSA images showing coil embolization of the aneurysm and preservation of the parent artery. H, I) DSA images at follow-up reexamination showing no evidence of aneurysm recurrence.

disease-associated distal AChA aneurysms that were treated by endovascular embolization and a review of the relevant literature summarizing the clinical features and treatment of this disease.

Case Reports

Case 1. A 38-year-old woman presented with an intraventricular hemorrhage. Physical examination revealed that she had no focal neuro-

Table 1 Clinical data for all cases of moyamoya disease-associated distal AChA aneurysms analyzed in this study.

Study	Year	Age/ sex	Presentation	Symptoms	Location	Treatment	Outcome
Tanaka et al. [3]	1980	57/F	IVH	Somnolence, hemiparesis	TH	Conservative + ventricular drainage	Death (IVH)
Furuse et al. [4]	1982	67/M	ICH, IVH	Confusion, hemiparesis	TH	Trapping (PA sac.) + excision	Stationary
Kasamo et al. [5]	1984	55/F	SAH (Acom aneurysm)	Meningeal irritation	TH	Acom aneurysm clipping + conservative for AChA aneurysm	Death (pneumonia)
Konishi et al. [6]	1985	18/F	ICH, IVH	Somnolence, flaccid hemiplegia	Lat. V	Conservative	Fair (dis. 4 mos.)
Konishi et al. [6]	1985	13/M	IVH	Vomiting, dist. cons.	Lat. V	Conservative	Fair (dis. 11 mos.)
Konishi et al. [6]	1985	57/F	IVH	Vomiting, dist. cons.	Lat. V	Conservative	Death (IVH)
Konishi et al. [6]	1985	34/F	IVH	Vomiting, dist. cons.	Lat. V	Conservative	Death (IVH)
Nakai et al. [7]	1992	46/F	ICH	Hemiparesis, dist. cons.	Trigone	Trapping (PA sac.) + excision	Stationary
Hamada et al. [8]	1994	48/F	IVH (rebleed 5 days later)	Headache, nausea, vomiting	Lat. V	Trapping (PA sac.) + STA-MCA anastomosis	Good
Kawai et al. [9]	1997	19/M	IVH	Headache	TH	Conservative	Fair (dis. 2 years)
Lee et al. [10]	2001	48/M	ICH, IVH	Somnolence, left hemiparesis	Trigone	Trapping (PA sac.) + excision	Good
Wong et al. [11]	2003	62/F	ICH	Disorientated, hemiplegia	TH	Clipping (PA pres.)	Good
Kim et al. [12]	2009	43/F	IVH	Stupor	Lat. V	nBCA embolization (PA sac.)	Stationary
Present case 1	2010	56/F	Headache	NONE	TH	Coiling embolization (PA pres.)	Good
Present case 2	2010	38/F	IVH, headache	NONE	Trigone	GLUBRAN embolization (PA sac.)	Good

Abbreviations: dist. cons., disturbed consciousness; Lat. V, lateral ventricle; PA, parent artery; pres., preserved; sac., sacrificed; dis., disappeared by.

logical signs. Brain computed tomography (CT) scan revealed hyperdense signs in bilateral lateral ventricles, which were especially prominent in the left trigone and temporal horn (Figure 1A). A cranial magnetic resonance imaging (MRI) scan revealed abnormal nodular isointense T1 and short T2 signals in the posterior horn of the left lateral ventricle. Subsequent digital subtraction angiography (DSA) revealed moyamoya-like vascular changes in both internal carotid arteries (Figure 1B-C). The left AChA was enlarged and extended to distal regions. An aneurysm 5 mm in diameter was noted at the distal end of the left anterior choroidal artery (Figure 1D-F). Therefore, the patient was diagnosed with a distal AChA an-

eurysm associated with moyamoya disease. Endovascular embolization of the aneurysm was performed under general anesthesia. A Marathon microcatheter was introduced into the proximal end of the aneurysm over a MirageTM 008 guidewire (Figure 1G). The aneurysm and the parent artery were then embolized with a Glubran 2 (GEM s.r.l., Viareggio, Italy)/lipiodol mixture (1:3). Subsequent angiography confirmed disappearance of the aneurysm (Figure 1H). The patient recovered well postoperatively. A one-year follow-up examination demonstrated that no rebleeding had occurred. Meanwhile, DSA reexamination showed no evidence of aneurysm recanalization and good collateral circulation (Figure 1I).

Figure 3 Schematic illustration of the location of all 13 cases of moyamoya disease-associated distal AChA aneurysms. Based on their precise locations, these aneurysms were divided into three groups: those in the temporal horn of the lateral ventricle, those in the lateral ventricle, and those in the trigone of the lateral ventricle. Therapeutic strategies employed included conservative treatment (black figures), craniotomy (red figures), and endovascular intervention (green figures).

Case 2. A 56-year-old woman presented with a five-month history of intermittent headache and dizziness. Physical examination revealed no focal neurological signs. CT angiography (CTA) revealed moyamoya-like vascular changes in both internal carotid arteries (Figure 2A). The left AChA was enlarged and extended to distal regions. An aneurysm-like lesion with a 4-mm diameter was noted in the trunk of the left AChA (Figure 2B-E). Subsequent DSA confirmed the initially suspected diagnosis of distal left AChA aneurysm associated with moyamoya disease.

Endovascular embolization of the aneurysm was performed under general anesthesia. An Echelon-10 microcatheter was introduced into the aneurysm cavity over a Traxcess-14 microguidewire. Following release of the Morpheus 3D CSR coil (4 mm × 10 cm), the MeproPlex 10 coil (2 mm × 3 cm) was deployed for embolization (Fig. 2F). Subsequent angiography revealed a patent parent artery and disappearance of the aneurysm (Figure 2G). The patient recovered well postoperatively. It was confirmed at a one-year follow-up examination that no rebleeding had occurred. DSA reexamination showed a patent parent artery and no evidence of aneurysm recanalization (Figure 2H, I).

Literature Review

A total of 15 cases of distal AChA aneurysms associated with moyamoya disease, including the two cases reported here and 13 identified through a MEDLINE search (1980-2010) were

analyzed. The clinical data for these patients are summarized in Table 1.

General data

Of the 15 patients included in the analysis, four were males and 11 were females. They ranged in age from 13 to 67 years, with a median age of 44.1 years. The types of onset included intracerebral hemorrhage (ICH) in two cases, intraventricular hemorrhage (IVH) in eight cases, IVH with ICH in three cases, headache and dizziness in one case, and subarachnoid hemorrhage (SAH) caused by rupture of another type of aneurysm in one case.

Aneurysm characteristics

DSA was performed to provide an accurate diagnosis in all cases. A ruptured aneurysm was diagnosed if the location of aneurysm overlapped with that of a hematoma. Most of the aneurysms (13/15) were ruptured, while just two were unruptured. The distal AChA aneurysms were located in the temporal horn of the lateral ventricle (six cases), in the lateral ventricle (six cases), and in the trigone of the lateral ventricle (three cases) (Figure 3).

Treatments

A conservative treatment course was followed in seven out of 15 patients. Meanwhile, five patients received craniotomy, including four who were subjected to aneurysm trapping and resection and one who underwent aneurysm clipping without sacrificing the parent artery. Among the four cases in which aneurysm trapping and resection was performed, there was parent artery sacrifice in three cases and

parent artery sacrifice with superficial temporal artery-middle cerebral artery (STA-MCA) anastomosis in one case. Additionally, three patients received endovascular therapy, including two who underwent embolization of the aneurysm and parent artery with liquid embolic agents [N-butyl cyanoacrylate (nBCA) or Glubran 2] and one who underwent coil embolization of the aneurysm with preservation of the parent artery (Figure 3).

Treatment outcome

Of the seven patients who followed a conservative treatment course, four died (three of bleeding and one of complicated pneumonia) and three showed a disappearance of the aneurysms. In eight cases, the patients' distal AChA aneurysms were subjected to aggressive treatment (craniotomy or endovascular intervention). The aggressively treated cases included three cases of aneurysms located in the temporal horn, of which one was treated by aneurysm trapping and resection and had sequelae of hemiparesis, one was treated by aneurysm clipping without sacrificing the parent artery resulting in a fair outcome, and one was treated by embolization of the aneurysm resulting in a good outcome.

In the two cases in which the aneurysms were located in the lateral ventricle, one was treated by aneurysm trapping and resection with STA-MCA anastomosis resulting in a good outcome, and one was treated by embolization of the aneurysm and parent artery with nBCA resulting in sequelae of hemiparesis. Of the three cases of aneurysms located in the trigone, two were treated by aneurysm trapping and resection. One of these patients had a good prognosis, while the other showed no symptom improvement postoperatively. The third case involving an aneurysm in the trigone was treated by embolization of the aneurysm and parent artery with Glubran 2 resulting in a good outcome.

Endovascular intervention

One of the three patients who received endovascular intervention had an aneurysm in the temporal horn and underwent coil embolization of the aneurysm with the parent artery preserved and exhibited a good prognosis. The second patient with an aneurysm in the lateral ventricle underwent nBCA embolization of the aneurysm and parent artery, and had the sequelae of hemiparesis. The third patient with an

aneurysm in the trigone, underwent Glubran 2 embolization of the aneurysm and parent artery, and showed a good prognosis.

Discussion

AChA aneurysms, occurring often at the junction of the internal carotid artery and the anterior choroidal artery, are rarely observed, and account for less than 5% of all intracranial aneurysms¹⁸. Distal AChA aneurysms are particularly rare. In a large surgical series of 1,012 patients with aneurysms, there was no patient reported with a distal AChA aneurysm¹⁹. Inci et al.² reported two cases of such aneurysms in 2007 and identified another 23 cases by performing a review of the 1955-2005 medical literature. Our search of the 2005-2010 MEDLINE literature identified only one case of a distal AChA aneurysm¹². Thus, a total of 28 cases of such aneurysms have been reported to date, indicating that the incidence rate of distal AChA aneurysms is very low. Of the 28 cases of AChA aneurysms identified, 13 (46.4%) were associated with moyamoya disease (Table 1), suggesting that the development of distal AChA aneurysms may be closely associated with moyamoya disease¹.

After originating from the internal carotid artery, the AChA proceeds posteriorly into the cistern and then passes through the choroidal fissure to reach the choroid plexus of the temporal horn. AChA branches extend to distal regions to reach the trigone of the lateral ventricle and form a rich collateral circulation network in the choroid plexus and lateral geniculate body by connecting to the posterior communicating artery, posterior cerebral artery, and posterior choroidal artery^{19,20}. On normal angiograms, the anterior choroidal artery is indiscernible within the choroid plexus²¹. When moyamoya disease occurs, because the internal carotid arteries are blocked, the trunk and branches of the AChA develop compensatory dilation, extend to distal regions, and form collateral vessels to increase blood flow. As a result, AChA branches are clearly visible on angiograms. It has been reported that in 73% of patients with moyamoya disease the AChA and its branches develop compensatory dilation and extend to distal regions to increase the blood supply. However, the increase in blood flow is associated with a high risk of development and rupture of distal AChA aneurysms¹.

After the AChA originates from the internal carotid artery, it extends to distal regions in a wavy pattern. The distal trunk of the AChA is called the distal AChA ²⁻¹². Using the choroid fissure as a boundary point, the distal AChA divides into two segments: intracisternal and intraplexual. On DSA images, this boundary point is also known as the ventral choroidal point ⁶. Of the 28 cases of distal AChA aneurysms identified thus far ^{2,12}, only two (7.1%), that presented with SAH, were located in the intracisternal segment, and the remaining 26 (92.9%), that presented with ICH or IVH, were located in the intraplexual segment. All 13 cases of moyamoya disease-associated distal AChA aneurysms were located in the intraplexual segment. Enlargement and distal extension of the intraplexual segment were clearly visible in DSA images (Figure 3).

The therapeutic strategies used for distal AChA aneurysms associated with moyamoya disease have varied over time. Conservative treatment was more often used prior to the maturation of microsurgical techniques. Seven of 13 patients diagnosed with moyamoya disease-associated distal AChA aneurysms underwent conservative treatment, of whom three showed aneurysm disappearance and four died shortly after disease onset. Because there have been so few reported cases of moyamoya disease-associated distal AChA aneurysms, little is known of their natural history. However, conservative treatment may be associated with a poor prognosis. In recent years, the presence of aneurysms has become recognized as a risk factor affecting the prognosis of moyamoya disease. More importantly, since moyamoya disease-associated distal AChA aneurysms are often located on collateral or moyamoya arteries, aneurysm rupture may lead to disastrous consequences ^{12,22,23}. Therefore, immediate and aggressive treatment should be given to patients with such aneurysms.

Surgical treatment and endovascular intervention are two main strategies used in the management of moyamoya disease-associated distal AChA aneurysms. The more frequent use of craniotomy (n=5) relative to endovascular intervention (n=3) may be due to the fact that endovascular techniques were developed after craniotomy. Unlike aneurysms of the circle of Willis, moyamoya disease-associated distal AChA aneurysms have no clear anatomic landmarks. Moreover, they are closely associated with the ventricular system. Consequently, it is

very difficult to identify and expose these aneurysms during craniotomy. Furthermore, craniotomy often causes great damage to nervous tissue and collateral circulation. More importantly, because intracranial arteries are often very vulnerable in moyamoya disease, the preservation of the parent artery is very difficult during craniotomy ^{13,24}. In contrast, DSA-guided endovascular intervention has the advantages of minimal invasiveness and accurate localization. With the continuous development of embolic agents and the advancement of technology, endovascular intervention has been applied widely in clinical practice ^{14,15}. A retrospective analysis of three cases of moyamoya disease-associated distal AChA aneurysms in which endovascular embolization was used indicated that the interventional approach was feasible. No significant differences were noted in preservation of the parental artery, postoperative complications, or prognosis between patients who underwent endovascular treatment and those who underwent surgical treatment. On the other hand, Inci et al. ² retrospectively analyzed 25 cases of distal AChA aneurysms and proposed that craniotomy is the preferred treatment for the disease. They suggested that endovascular embolization may affect the proximity of the AChA, and that reflux of the liquid embolic agents may result in disastrous consequences. However, the presence of new embolic agents and the application of catheter techniques in recent years have enabled endovascular treatment to become a more effective and safer approach ²⁵⁻²⁶. The observation that good outcomes were achieved in the 2 cases reported here after endovascular therapy provides further supportive evidence for the efficacy of this approach.

Our analysis of the clinical data of eight cases of moyamoya disease-associated distal AChA aneurysms undergoing aggressive treatment suggested that aneurysm location and preservation of the parent artery are two major factors affecting the prognosis of the disease. In contrast, different therapeutic strategies (craniotomy or endovascular intervention) appeared to have no significant impact on the prognosis of the disease.

Marincovic et al. ²⁷ pointed out that embolization of the intraplexual segment is safe, since the main branches of the AChA have diverged before it enters the temporal horn of the lateral ventricle. However, Fujii et al. ²⁸ and Morandi et al. ²⁹ have suggested that embolization of the

intraplexual segment may cause ischemic symptoms since the distal AChA has branches that supply the thalamus. In all 13 cases of moyamoya disease-associated distal AChA aneurysms identified in this paper, enlargement and distal extension of the AChA and formation of rich collateral circulation were noted on DSA images. Therefore, we suggest that embolization of the intraplexual segment can cause ischemia. The eight cases of moyamoya disease-associated distal AChA aneurysms that were treated aggressively were further divided into three groups based on the precise locations of the aneurysms: those in the temporal horn of the lateral ventricle (n=3), those in the lateral ventricle (n=2), and those in the trigone of the lateral ventricle (n=3). Regardless of whether surgical treatment or endovascular embolization was used, preservation of the parent artery was often associated with a favorable postoperative outcome. In the three cases of aneurysms located in the trigone of the lateral ventricle, simultaneous embolization of the aneurysm and the parent artery did not cause new postoperative complications, which suggests that the parent artery can be sacrificed when aneurysms are located in the distal end of the AChA.

There are two types of AChA aneurysm associated with moyamoya disease^{1,4,6,12}. One is true aneurysm which develops by increasing flow to distal portion, with or without hemorrhagic insult. The other is pseudoaneurysm associated with hemorrhagic insult which contains no structural elements of arterial wall, usually located at the distal end of the AChA or in the collateral circulation. Each of the two types of aneurysm requires different management strategy at the time of endovascular treat-

ment. In pseudoaneurysm, endovascular treatment of aneurysm space itself such as endovascular coil embolization does not make sense to prevent future re-hemorrhage, and the orifice of pseudoaneurysm in the parent artery itself should be eliminated by small amount of glue or coils. Considering distal collateral flow, glued normal vessel should be minimal to keep distal collateral flow^{4,6}. On the other hand, flow-related true aneurysm could be treated by endovascular coil embolization if possible, and the parent artery should be preserved to achieve better outcome. In Case 1 reported here, the aneurysm located at the ending of the AChA belongs to a pseudoaneurysm and was treated by parent artery occlusion. In contrast, the aneurysm located in the trunk of the left AChA in Case 2 represents a true aneurysm and was managed by coil embolization with the parent artery preserved. A good outcome was achieved in both cases.

Conclusions

Our findings suggest endovascular embolization is an effective and feasible treatment for distal AChA aneurysms associated with moyamoya disease. Aneurysm location and preservation of the parent artery are two major prognostic factors for patients with moyamoya disease-associated distal AChA aneurysms undergoing either craniotomy or endovascular intervention. To achieve better treatment outcomes, the parent artery should be preserved when the aneurysm is located in the temporal horn of the lateral ventricle, but can be sacrificed when the aneurysm is located in the trigone of the lateral ventricle.

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