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Safety and effects of endovascular treatment of basilar tip aneurysms in patients with moyamoya diseases

Chun-Guang Chen, MD, PhD[a], Bu-Lang Gao, MD, PhD[b,*], Cheng-Bao Yang, MD[b], Xiao-Hong Hao, MD[b], Chun-Feng Ren, MD, PhD[c], Lei Yang, MD, PhD[b], Yong-Feng Han, MD, PhD[b], Qin-Ying Cao, MD, PhD[b]

Abstract

The effect and safety of endovascular treatment of basilar tip aneurysms associated with moyamoya disease are unknown. This study was to investigate the safety and effect of endovascular treatment of basilar tip aneurysms associated with moyamoya disease. Patients with moyamoya disease concurrent with basilar tip aneurysms were retrospectively enrolled and treated with endovascular embolization. The clinical and angiographic data were analyzed. Thirty patients with a basilar tip aneurysm were enrolled, including 8 (26.67%) male and 22 (73.33%) female patients aged 38 to 72 years (mean 54.4 ± 8.15). Endovascular treatment was successfully performed in 29 (96.67%) patients but failed in 1 (3.33%). Immediately after embolization, aneurysm occlusion degree was Raymond-Roy grade I in 26 (89.66%), grade II in 2 (6.90%), and grade III in 1 (3.45%). Intraprocedural complications occurred in 2 (10%) patients, including aneurysm rupture in 1 (3.33%), leading to death of the patient, and stent thrombosis in 2 (6.67%) which was successfully treated with thrombolysis. At discharge, good clinical outcome (modified Rankin Scale 0–2) was achieved in 29 (96.67%) and death in 1 (3.03%). Follow-up was performed 6 to 26 months (median 15) in 27 (93.1%) patients. Aneurysm occlusion degree was Raymond-Roy grade I in 21 (77.78%) patients, grade II in 4 (14.81%), and grade III in 2 (7.41%), not significantly (P = .67) different from those immediately after embolization. Aneurysm recurrence was found in 4 patients (14.81%). The clinical outcome was modified Rankin Scale 0 to 2 in all 27 patients, not significantly different from that at discharge. Endovascular embolization can be performed safely and effectively for basilar tip aneurysms associated with moyamoya disease even though more advanced embolization techniques are necessary.

Abbreviation: mRS = modified Rankin Scale.

Keywords: basilar tip, cerebral aneurysms, endovascular, moyamoya disease, stent-assisted coiling

1. Introduction

As a rare cerebrovascular disease, moyamoya disease is characterized by a special vascular network at the base of the brain caused by increasingly-severe stenosis and occlusion of the supraclinoid internal carotid artery and the major arterial branches within the Willis circle.[1–4] Cerebral aneurysms may accompany moyamoya disease occasionally, and the incidence of accompanied cerebral aneurysms is estimated to range from 3% to 14%.[5] These aneurysms may be classified into the peripheral and major artery aneurysms. The peripheral aneurysms originate primarily from distal fragile arteries and may easily rupture to cause subarachnoid hemorrhage, whereas the major trunk aneurysms usually result from hemodynamic disturbance and may have a higher risk of spontaneous rupture.[5] The prognosis of rupture of these accompanying aneurysms is relatively poor, and early intervention of these aneurysms in patients with moyamoya disease is necessary to improve the prognosis. Basilar tip aneurysms associated with moyamoya disease have been reported infrequently,[5–9] with most studies reporting only few cases. It was hypothesized that endovascular embolization could be performed safely and effectively for basilar tip aneurysms associated with moyamoya disease. This study was to investigate the safety and effect of endovascular treatment of basilar tip aneurysms in patients with moyamoya disease.

2. Materials and methods

This retrospective study was approved by the ethics committee of Shijiazhuang People Hospital, and all patients had given

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The datasets generated during and/or analyzed during the current study are not publicly available, but are available from the corresponding author on reasonable request.

[a] Department of Neurosurgery, Liaoyang City Central Hospital, Liaoyang City, Liaoning Province, China, [b] Department of Neurosurgery, Shijiazhuang People's Hospital, Liaoyang City, Liaoning Province, China, [c] Department of Laboratory Analysis, Zhengzhou University First Affiliated Hospital, Liaoyang City, Liaoning Province, China.

** Correspondence: Bu-Lang Gao, Department of Neurosurgery, Shijiazhuang People's Hospital, 365 South Jianhua Street, Shijiazhuang, Hebei Province 050011, China (e-mail: browngao@163.com).*

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their signed informed consent to participate. Between 2014 and 2020, patients with moyamoya disease concurrent with basilar tip aneurysms which were treated endovascularly were retrospectively enrolled. The inclusion criteria were patients with moyamoya disease which was confirmed by cerebral angiography, concurrent with basilar tip aneurysms treated with endovascular embolization. The exclusion criteria were patients who were allergic to contrast agent and patients with severe heart, liver and kidney diseases which prevented them from being treated surgically.

Three to 5 days before the embolization procedure, dual antiplatelet therapy with clopidogrel (75 mg/d) and aspirin (100 mg/d) was administered. The endovascular procedure was performed under general anesthesia. After femoral access, systematic heparinization was performed with an intravenous bolus injection of heparin at 70 to 100 U/kg followed by continuous infusion of heparin so that an activated clotting time of 2.5 to 3 times to their baseline activated clotting time was maintain during the procedure. A guiding catheter was inserted for cerebral angiography to evaluate the cerebral vasculature, size and location of the basilar tip aneurysms. Then, under roadmap guidance, a microcatheter was navigated to the aneurysm sac for coiling alone and coiling with assistance of stents or 2 microcatheters. For stent-assisted coiling, the patients were treated with dual antiplatelet medications (100 mg aspirin and 75 mg clopidogrel) for 6 weeks and then aspirin 100 mg for 6 months after embolization. Follow-up was performed 6 to 12 months after embolization.

Aneurysm size and location, treatment modality using coils or stents, immediate aneurysm occlusion degree after embolization, intraprocedural complications, clinical and angiographic outcomes at discharge and follow-up were assessed. Aneurysm occlusion degrees were evaluated using the Raymond–Roy grading system, with grade I for complete occlusion, grade II for neck remnant, and grade III for aneurysm sac remnant.[10] The clinical outcome was assess with the modified Rankin Scale (mRS) score, with good clinical outcomes as mRS 0 to 2 and poor as mRS 3 to 6.

2.1. Statistical analysis

The statistical analysis was performed with the SPSS software (version 19.0, IBM, Chicago, IL). Measurement data were presented as mean ± standard deviation if in normal distribution but as median and interquartile range if in skew distribution. Enumeration data were presented in frequency and percentages. For measurement data with normal distribution, the Student t test was used to detect the difference between 2 groups, and for continuous measurement data with skew distribution, the Mann–Whitney *U* test was used to test the difference between groups. Enumeration data were tested using the Chi square test or Fisher exact test. The significant *P* value was set at <.05.

3. Results

Thirty patients with a basilar tip aneurysm were enrolled, including 8 (26.67%) male and 22 (73.33%) female patients with an age range of 38 to 72 years (mean 54.4 ± 8.15) (Table 1). The size of the aneurysm ranged 4 to 16 mm (mean 8.20 ± 3.32), with 22 (73.33%) small aneurysms (3–10 mm in size) and 8 (26.67%) large ones (11–16 mm in size). The initial symptom was transient ischemic attack in 13 (43.33%) patients, subarachnoid hemorrhage caused by aneurysm rupture in 5 (16.67%), headache in 4 (13.33%) patients, and cerebral infarction in 2 (6.67%). The other aneurysms in 6 (20%) patients were incidentally found.

Endovascular treatment was successfully performed in 29 (96.67%) patients but failed in 1 (3.33%) because of severe vasospasm which prevented access of the aneurysm. Coiling alone was performed in 1 patient (0.33%), coiling with double microcatheters in 9 (30%), stent-assisted coiling with 2 stents in 3 (10%) patients, and stent-assisted coiling using 1 stent in 16 (53.33%). In 5 patients with subarachnoid hemorrhage caused by aneurysm rupture, 3 patients were treated in the acute stage using 2 microcatheters for coiling in 2 and stent-assisted coiling in the other 1. The other 2 patients were treated with stent-assisted coiling in 1 and coiling alone in the other 1 beyond 1 month after aneurysm rupture. Twenty-two stents were deployed, including the Enterprise stent (n = 8), Solitaire EZ (n = 6), and LVIS (n = 8).

Immediately after embolization, aneurysm occlusion degree was Raymond–Roy grade I in 26 (89.66%), grade II in 2 (6.90%), and grade III in 1 (3.45%) (Table 1).

Intraprocedural complications occurred in 3 (10%) patients, including aneurysm rupture in 1 (3.33%) patient with a large aneurysm (13 mm in diameter), leading to death of the patient, and stent thrombosis in 2 (6.67%, with 1 large aneurysm of 15 mm in size and the other small aneurysm of 10 mm in size) which was successfully treated with thrombolysis (10 mL tirofiban). At discharge, good clinical outcome (mRS 0 to 2) was achieved in 29 (96.67%) and death in 1 (3.03%).

Follow-up was performed 6 to 26 months (median 15) in 27 (93.1%) patients, with 2 patients being lost. Aneurysm occlusion degree was Raymond–Roy grade I in 21 (77.78%) patients, grade II in 4 (14.81%), and grade III in 2 (7.41%), which were not significantly (*P* = .67) different from those immediately after embolization. Aneurysm recurrence was found in 4 patients (14.81%) with large aneurysms (over 10 mm in size), and no retreatment was performed. The clinical outcome was mRS 0 to 2 in all 27 patients, not significantly different from that at discharge.

4. Discussion

In this study, investigating the effect and safety of endovascular treatment for basilar tip aneurysms associated with moyamoya disease, endovascular embolization could be performed safely and effectively for basilar tip aneurysms associated with moyamoya disease even though more advanced embolization techniques are necessary.

Cerebral aneurysms associated with moyamoya disease are dangerous because of a high risk of rupture. A rupture rate of 89% has been reported in 111 patients of cerebral aneurysms associated with moyamoya disease by Kawaguchi et al,[11] and in the study by Ni et al[5] including 34 patients of cerebral aneurysms associated with moyamoya disease, 32 (94.12%) patients presented with varied types of intracranial hemorrhage, indicating the high risk of hemorrhage of these kinds of cerebral aneurysms. This necessitates immediate and effective treatment of these aneurysms. Although these aneurysms have been reported in the literature, the studies were only limited to a small number of cases and case reports,[1–9] which were not sufficient to develop an optimal therapeutic strategy.

For peripheral aneurysms developed on the moyamoya vessels and collateral vessels, direct surgical interventions like aneurysmectomy or neck clipping could be effective. Nonetheless, deep and distal location and fragile moyamoya and perforating vessels may hinder the surgery or cause aneurysm rupture, which may prevent surgical intervention in these patients. Endovascular embolization is less invasive and may be the first choice of treatment for these peripheral aneurysms, however, the endovascular approach may be limited if the parent artery is tortuous. Revascularization with bypass surgery may be effective, and by developing additional collateral flow in the surgical territory and decreasing the hemodynamic stresses on the pathological vessels of aneurysms, the distal aneurysms may shrink. Ni et al has reported a 92.3% (12/13) obliteration rate of peripheral aneurysms using the revascularization bypass

Table 1

Patients' data, treatment, and follow-up outcome.

Variables		Data
No. of patients		30
F/M		22/8
Age (y)		8–72 (mean 54.4 ± 8.15)
Symptoms	Transient ischemic attack	13 (43.33%)
	Aneurysm rupture	5 (16.67%)
	Headache	4 (13.33%)
	Cerebral infarction	2 (6.67%)
	Incidentally found	6 (20%)
Aneurysm size (mm)	4–16 (mean 8.20 ± 3.32)	
	<3	0 (0%)
	≥3 and ≤ 10	22 (73.33%)
	>11 and ≤ 16	8 (26.67%)
	>16	0 (0%)
Endovascular treatment	Coiling alone	1 (3.33%)
	Coiling with 2 microcatheters	9 (30%)
	Stent-assisted coiling with two stents	3 (10%)
	Stent-assisted coiling with one stent	16 (53.33%)
	Failure of embolization	1 (3.33%)
Immediate aneurysm occlusion	Raymond–Roy grade I	26 (89.66%)
	Grade II	2 (6.90%)
	Grade III	1 (3.45%)
Intraprocedural complications	Aneurysm rupture	1 (3.33%)
	Stent thrombosis	2 (6.67%)
Outcome at discharge	mRS 0–2	29 (96.67%)
	mRS 6 (death)	1 (3.03%)
Follow-up	Time	6–26 (median 15)
	Patients	27 (93.1%)
Aneurysm occlusion degree	Raymond–Roy grade I	21 (77.78%)
	Grade II	4 (14.81%)
	Grade III	2 (7.41%)
	Aneurysm recurrence	4 (14.81%)
Outcome at follow-up	mRS 0–2	27 (100%)

mRS = modified Rankin Scale score.

approach, which may strengthen the relationship between the formation of peripheral aneurysms and hemodynamic stresses in moyamoya disease.[5]

For main trunk aneurysms located in the Willis circle, persistent high-flow stresses may be the cause of formation of these aneurysms because of blood compensation on the vertebrobasilar system in the case of occlusion of the internal carotid artery.[1,12,13] Direct surgical intervention may generally leads to good clinical outcomes in these aneurysms.[14] Revascularization with bypass surgery may alter the hemodynamic stresses associated with moyamoya disease, leading to shrinkage of the posterior circulation aneurysms.[5] This is because revascularization may reconstruct the anterior circulation to relieve the blood flow burden on the vertebrobasilar system.

Endovascular embolization may be an alternative choice for these aneurysms in the posterior circulation. Nonetheless, stent-assisted coiling is not recommended for these aneurysms because of the antiplatelet therapy for stent deployment, especially in case of aneurysm rupture.[5] For posterior circulation aneurysms, coil embolization has become the preferred treatment of choice because of the difficulty and hazards of direct surgical clipping.[11,15,16] However, coil embolization without use of stents is only applicable to aneurysms with a narrowed neck, and for wide-necked aneurysms, stent-assisted coiling is necessary to prevent coil protrusion and escape and increase dense embolization. In our study with 30 basilar tip aneurysms, stent-assisted coiling was performed in 19 (63.33%) patients, with 2 stents used in 3 (10%) patients and 1 stent in the other 16 (53.33%). In ten (33.33%) patients treated with coiling embolization without stent assistance, 2 microcatheters were used in 9 (30%) patients, and 1 microcatheter in only 1 (3.33%) patient. Thus, 28 (93.33%) patients were treated with stent-assisted coiling or 2 microcatheters for coiling. The use of advanced embolizing technique may be caused by strong blood flow in the basilar artery caused by blood compensation in moyamoya disease. The strong blood flow makes endovascular devices unable to stay stably, and advanced techniques have to be applied to densely pack the aneurysm cavity. Under direct impact of strong blood flow, the densely packed aneurysms may easily recur, which may be the reason for aneurysm recurrence at follow-up. In our study, aneurysm recurrence was found in 4 patients (14.81%) during follow-up 6 to 26 (median 15) months after embolization. It has been reported that endovascular embolization of cerebral aneurysms has a higher recurrence rate, ranging 15% to 33%.[17–19] The recurrence rate in our study seemed to be consistent with this range of recurrence rate, nonetheless, stent-assisted coiling may reconstruct the parent artery blood flow and the hemodynamic stresses with the stent struts, thus decreasing the impact of strong blood flow on the coils within the aneurysm cavity.

In our study, the aneurysm ruptured in 5 patients, however, only 3 patients were treated in the acute stage, using 2 microcatheters for coiling in 2 and stent-assisted coiling in the other 1. The other 2 patients were treated beyond 1 month after aneurysm rupture. Although antiplatelet therapy may increase bleeding risk for aneurysms associated with moyamoya disease, especially in case of subarachnoid hemorrhage caused by aneurysm rupture, the 3 cases treated at the acute stage of aneurysm rupture did not show any adverse effects, including 1 case with stent thrombosis which was treated with thrombolysis. However, more intense studies are necessary to investigate the risk of ruptured aneurysms treated with stent-assisted coiling.

Some limitations existed in this study, including 1-center and retrospective study design, a small cohort of patients, Chinese patients enrolled only, and a short period of follow-up. All these issues may affect the generalization of the outcomes which should be explained with caution. Future studies overcoming all these issues should be carried out for better outcomes.

In conclusion, endovascular embolization can be performed safely and effectively for basilar tip aneurysms associated with moyamoya disease even though more advanced embolization techniques are necessary.

Author contributions

Conceptualization: Chun-Guang Chen, Bu-Lang Gao, Lei Yang, Qin-Ying Cao.
Data curation: Chun-Guang Chen, Xiao-Hong Hao, Chun-Feng Ren, Lei Yang, Yong-Feng Han.
Formal analysis: Chun-Guang Chen, Bu-Lang Gao, Cheng-Bao Yang, Qin-Ying Cao.
Funding acquisition: Bu-Lang Gao, Xiao-Hong Hao, Lei Yang, Qin-Ying Cao.
Investigation: Bu-Lang Gao, Cheng-Bao Yang, Yong-Feng Han.
Methodology: Chun-Guang Chen, Xiao-Hong Hao, Chun-Feng Ren, Yong-Feng Han.
Resources: Cheng-Bao Yang.
Supervision: Xiao-Hong Hao, Lei Yang, Qin-Ying Cao.
Validation: Chun-Guang Chen, Bu-Lang Gao, Cheng-Bao Yang, Xiao-Hong Hao, Chun-Feng Ren, Lei Yang, Yong-Feng Han, Qin-Ying Cao.
Visualization: Chun-Guang Chen, Bu-Lang Gao, Chun-Feng Ren, Yong-Feng Han.
Writing – original draft: Bu-Lang Gao.
Writing – review & editing: Bu-Lang Gao.

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