Peripheral aneurysms in the lateral posterior choroidal artery (LPChA) are rare (1-5). Open surgical treatment requires a transcortical approach to the lateral ventricle, which is difficult and associated with a rather high morbidity and mortality rate (6). Similarly, endovascular treatment with controllable coils can also be difficult or impossible due to the distal location of the aneurysm or an unfavorable ratio between the size of the aneurysm and the size of the parent artery.

This report describes a case of moyamoya disease with a thalamic hematoma and aneurysm in the distal LPChA unassociated with the thalamic hemorrhage. Endovascular treatment with glue was performed, resulting in an excellent anatomical and clinical outcome.

**Key Words:** Choroidal artery aneurysm; Moyamoya disease; Treatment

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**CASE REPORT**

A 45-year old female presented with a sudden onset of severe headaches and dysarthria, plus a history of an ischemic stroke 18 months previously. A computed tomographic scan of the head revealed a right thalamic hemorrhage and old infarction in the left frontal area (Fig. 1). For further evaluation, magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) were performed, where the latter clearly revealed stenosis and/or occlusion in the terminal portion of the internal carotid artery and proximal portion of the anterior and middle cerebral arteries. A 3-mm aneurysm arising from the distal portion of the right LPChA was also noted. The MRI and MRA both showed the right thalamic hematoma at a distance from the aneurysm (Fig. 2). A subsequent cerebral angiogram revealed stenosis and/or occlusion of the terminal portion of the bilateral internal carotid artery with moyamoya vesseles (Fig. 3A, 3B), plus a 3-mm
aneurysm in the choroidal portion of the right LPChA and moyamoya-like collaterals in the distal territory of the artery (Fig. 3C, 3D). After considering the therapeutic options, the patient agreed to endovascular treatment of the aneurysm 5 days later.

The endovascular treatment was performed under local anesthesia and systemic heparinization, and the adequacy of the systemic anticoagulation monitored by frequent measurements of the activated clotting time. A 6F Envoy guiding catheter (Cordis, Miami Lakes, FL)
was placed into the distal cervical left vertebral artery, and a Prowler 10 microcatheter (Cordis, Miami Lakes, FL) advanced over an Agility 0.010-inch microguidewire (Cordis, Miami Lakes, FL) into the distal right LPChA. The resulting superselective angiogram revealed a 3-mm aneurysm in the distal choroidal portion of the right LPChA and moyamoya-like collaterals in the distal territory of the artery, where the parietal portion of the right cerebral hemisphere was supplied by moyamoya-like vesseles arising from the distal portion to the aneurysm (Fig. 4A). Vertebral angiography performed with the microcatheter in a wedged state (total occluded state) in the distal LPChA revealed a good collateral flow in the hemispheric area supplied by moyamoya-like vesseles and retrograde filling of the aneurysm sac.

The aneurysm and just proximal parent artery to the aneurysm were then embolized carefully under fluoroscopic observation with a 1:3 mixture of n-butylecynoacrlyate (Histoacryl, B Braun, Melsungen, Germany) and iodized oil (Lipiodol, Andre Guerbert, Aulnay sous Bois, France). A control angiogram performed immediately after the embolization demonstrated successful occlusion of the distal LPChA and aneurysm (Fig. 4B, 4C, 4D). The patient recovered without any new neurological deficit, and was transferred to another hospital near her home 1 day later.

**DISCUSSION**

Peripheral aneurysms arising in the distal portions of both the supratentorial and infratentorial intracranial arteries are rare (7). To date, existing literature only contains 10 reported cases of aneurysms arising in the LPChA (1-5, 8, 9), all of which were small (<5 mm in diameter) and located distally. Seven of the ten LPChA aneurysms were associated with moyamoya disease (1-3, 5), one was associated with an arteriovenous malformation (4), and two were unassociated with moyamoya disease or arteriovenous malformation (9).

The high association rate of LPChA aneurysms with moyamoya disease suggests that patients with anterior circulation occlusive disease may be at greater risk of aneurysm formation, as in such cases the LPChA
typically functions as a high-flow collateral from the posterior circulation, putting it under greater hemodynamic stress. In the present case, stenosis and/or occlusion was found in the terminal portion of both the internal carotid artery and the proximal portion of the anterior and middle cerebral arteries, which is compatible with moyamoya disease.

Moyamoya disease most frequently presents with either ischemia or a hemorrhage (10, 11), where bleeding seems to occur primarily by two mechanisms: either rupture of the associated aneurysms or rupture of the collateral vessels exposed to unusual hemodynamic stress (12). The bleeding rate of aneurysms associated with moyamoya disease has been estimated to be 87.1% in adults (13). In the present case, the source of the hemorrhage was believed to be a rupture of a collateral vessel rather than an aneurysmal rupture originating in the right LPChA. The MRI and MRA showed that the thalamic hematoma occurred at a distance from the aneurysm, and no aneurysm or arteriovenous malformation was found adjacent to or within the thalamic hematoma. The aneurysm arising in the LPChA was located adjacent to the lateral ventricle wall, and thus was found incidentally.

The treatment strategy for such a case can be more complex. Open surgical treatment of LPChA aneurysms using a parieto-occipital craniotomy and transcortical-transventricular approach has already been reported in two cases (1, 4), where the aneurysm was trapped and the parent artery excised. However, both patients experienced adverse neurological sequelae, including transient exacerbation of a preexistent hemiparesis in one case (4), and the occurrence of a new hemiparesis in the other (1). In a recent article, Ali et al (9) described open surgical treatment of an LPChA aneurysm using frameless stereotactic guidance, the patient was returned to his baseline with known significant deficits. In their discussion, the authors pointed out the unique challenges of planning surgical intervention in patients with moyamoya disease and reemphasized that a surgical approach must minimize the risk of interrupting the rich collateral circulation that develops in these patients.

Thus, an endovascular approach is an attractive alternative, as it avoids the risk of interrupting the collateral circulation with a craniotomy and transcortical dissection. Nonetheless, even though endovascular coiling with parent artery preservation is generally preferable to occlusion of the parent artery, it may be difficult or impossible due to the distal location of the aneurysm or an unfavorable ratio between the size of the aneurysm and the size of the parent artery.

The concept of using glue agents, such as n-butylcyanoacrylate, to occlude an aneurysm or parent artery in the case of symptomatic distal aneurysms in supratentorial and infratentorial arteries is not new. However, there have only been three reports on the use of endovascular therapeutic occlusion for aneurysms arising in the LPChA (3, 8). In one of these reports, a ruptured aneurysm and the distal LPChA itself were embolized with n-butylcyanoacrylate in a patient with moyamoya disease (3). Unfortunately, the case was complicated by a large hemispheric infarct that extended well beyond the typical LPChA territory, and it was speculated that this artery played a critical role in providing collateral hemispheric blood flow. The other two reported cases were not associated with moyamoya disease, and the two symptomatic aneurysms were managed successfully using n-butylcyanoacrylate with excellent technical and clinical outcomes (8). In all three cases, the aneurysms were ruptured. Therefore, the present report represents a new (4th) case of endovascular management of a peripheral LPChA aneurysm that was found incidentally. As such, the endovascular treatment of an unruptured aneurysm in the distal LPChA has not yet been reported.

Superselective microcatheterization of the distal portion of the LPChA is usually technically possible with a microcatheter, although smaller systems are needed to safely navigate distally. Superselective angiography of the LPChA is essential before embolization is attempted, as it permits a more detailed analysis of the arterial microanatomy and a clearer delineation of the normal arterial supply derived from the LPChA to important diencephalic and mesencephalic structures than is possible from routine catheter angiography of the vertebral artery. The distal artery (intraventricular segment) primarily supplies the choroid plexus (14). In addition, the anterior and lateral posterior choroidal arteries form an anastomosis, limiting the risk of a symptomatic infarct when the distal artery is embolized (14). Clearly, occlusion of the proximal (intracisternal) segment of the LPChA is more likely to cause symptomatic ischemic injuries (14, 15). In moyamoya disease, the role of the LPChA is different from normal vascular anatomy, as the artery may play a critical role in providing collateral hemispheric blood flow (3). Thus, before embolization, provocative functional testing of an awake patient with a low Hunt and Hess grade may be helpful (16). In the present case, instead of a provocative functional test, vertebral angiography was performed with the
microcatheter in a wedged state (total occluded state) in the distal LPChA. Plus, good collateral flow in the hemispheric area supplied by moyamoya-like vessels and retrograde filling of the aneurysm sac were both observed.

n-butylcyanoacrylate was selected as the preferred embolic agent to ensure permanent occlusion of the aneurysm and just proximal parent artery, using a slow, well-controlled injection of the liquid adhesive. Owing to the fluidity of the adhesive mixture, this procedure has the advantage of permitting good downstream permeation of the targeted vascular segment (which includes the aneurysm), as well as complete flow control via a wedged injection (17). The polymerization rate of the embolic mixture must be adjusted to allow for this distal permeation, which was accomplished using a high ratio of iodinized oil to cyanoacrylate.

Aneurysms in the distal LPChA are exceedingly rare. The treatment strategy for an unruptured aneurysm, as in the present case, may be more complex. Thus, occlusion of an intracranial aneurysm in the distal LPChA and distal parent artery by injection of glue may be a safe and efficient alternative to surgery and endovascular therapy with controllable coils.

References
Endovascular Management of Peripheral Lateral Posterior Choroidal Artery Aneurysm Associated with Moyamoya disease

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