Two Different Methods of Endovascular Treatment for Ruptured Intracranial Aneurysm Associated with Moyamoya Disease and Review of the Literature

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Abstract: The purpose of this study was to evaluate efficacy and feasibility of two different embolization methods for the treatment of intracranial aneurysm with moyamoya disease. Two intracranial aneurysms with moyamoya disease treated with coils embolization and glue embolization respectively between September 2006 and December 2010 were analyzed and the related literatures were reviewed as well. The two intracranial aneurysms were successfully embolized and no complication of endovascular therapy occurred. We think that endovascular treatment may be a safe and efficacious method for the intracranial aneurysm with moyamoya disease, if coil embolization is difficult for some aneurysm, glue embolization may be a choice.

Keywords: Endovascular treatment; intracranial aneurysm; moyamoya disease

1. Introduction

The incidence of intracranial aneurysm (ICA) in adult patients with moyamoya disease has been estimated at 3%–14% (Borota et al, 1996). In contrast to the general population, ruptured ICA in moyamoya disease patients has a poorer prognosis (Kwak et al, 1984). Therefore, prevention of rebleeding from ruptured moyamoya related-ICA is of importance. There are two distinct options of treatment for ruptured ICA associated with moyamoya disease: direct surgical clip and endovascular embolization. The former is a standard approach, however, which is often difficult due to complicated moyamoya-like vessels in the operative field (Michael et al, 2009), moreover, moyamoya-like vessels are fragile and easy to rupture (Nishlo et al, 2004). The latter can avoid damaging brain tissue and such vessels (Suzuki et al, 2006). We report our successful experience in the treatment of ruptured ICA associated with moyamoya disease using two endovascular methods (detachable coil embolization and “glue” embolization). To the best of our knowledge, this is the first case report introducing two different endovascular methods treating ruptured ICA associated with moyamoya disease simultaneously.

2. Material and Methods

Case 1

A 52-year-old man presented with severe headache and vomiting for eight hours. On admission, he was fully conscious without focal signs. His condition was categorized as Hunt and Hess grade 1. Head CT showed subarachnoid hemorrhage in suprasellar cistern and cisterna ambiens (Fig 1-A). Cerebral angiography performed on the same day demonstrated occlusion in the terminal portion of bilateral internal carotid artery with moyamoya vessels and a 3.5-mm saccular ICA located on the left P2 segment of posterior cerebral artery (Fig 1-B, C.D.G.H). Following induction of general anesthesia, a 6F sheath was inserted in the right femoral artery and a 6F guiding catheter (Envoy; Cordis USA) was placed in the right vertebral artery. The patient was heparinized to an activated clotting time of 250 to 300 seconds. An Excelsior 10 microcatheter (Target/Boston Scientific) was navigated into aneurysm lumen. The aneurysm was then embolized with GDC-10 (3mm × 40mm, 2mm × 20mm, 2mm × 10mm Target/Boston Scientific USA). Angiography after coil embolization demonstrated occlusion of the body and dome of the aneurysm with excellent blood flow through left posterior cerebral artery. (Fig 1-E, F). No change in the patient’s neurological status was noted after the procedure and recovered in good condition.

Case 2

A 20-year-old man who presented with complaints of sudden, severe headache, nausea, and vomiting was transferred to our hospital after a diagnosis of lateral cerebral ventricle hemorrhage at an outside emergency department. He was fully conscious with left hemiparesis and was categorized as Hunt and...
Hess grade 2. Head CT revealed right lateral cerebral ventricle hemorrhage (Fig 2-A). Cerebral angiography revealed severe stenosis of bilateral internal carotid artery with moyamoya vessels and a 3-mm aneurysm located in a branch of right lateral posterior choroidal artery (LPChA) (Fig 2-B, C). On the day after the occurrence of bleeding, endovascular treatment was administered while the patient was under general anesthesia. Using the above method, an Excelsior 10 microcatheter was placed in the right lateral posterior choroidal artery over a 0.010-inch Transcend EX microguidewire (Target/Boston Scientific USA); however, the microcatheter could not catheterize the parent branching vessel. Then this vessel was catheterized by using a 1.3 F floating microcatheter (Marathon, ev3 USA). Superselective angiogram confirmed the ICA (Fig 2-D). 0.5 mL of a 7:1 mixture (at concentration of 12.5%) of iodized oil (Cordis Neurovascular) and cyanoacrylate glue (Glubran 2, GEM Italy) was injected through the microcatheter for 5 seconds under roadmap. Filling both the aneurysm and the adjacent parent artery with the cyanoacrylate glue, the microcatheter was then aspirated and quickly withdrawn. A control left vertebral artery angiogram demonstrated complete embolization of the aneurysm and the parent vessel (Fig 2-E). The patient remained neurologically unchanged and recovered in good condition.

3. Discussions

About half of adult patients with moyamoya disease develop intracranial bleeding, due to rupture of moyamoya-like vessels or ICA (Satoh et al, 2008). Currently, the formation of a moyamoya-related ICA is generally believed to be result of the increased wall stress due to high flow imposed by collateral circulation or anomalous arteriovenous shunt (Dietrichs et al, 1992).

In general, the formation of a moyamoya-related ICA is located in the posterior cerebral circulation, whereas in moyamoya disease patients, this proportion is 43.3% (Murakami et al, 2004). In moyamoya disease, occlusive or serious stenosis of distal segment of internal carotid artery and proximal segment of middle cerebral artery may decrease the flow dynamics across anterior cerebral circulation which increase flow through posterior cerebral circulation and originate turbulence that increase possibility of ICA formation and rupture. In this report, both ICAs located in the posterior cerebral circulation.

There are two treatment options available to moyamoya-related ICA: craniotomy with clip ligation (clipping) and endovascular embolization with detachable coils (coiling) or liquid embolic agent (glue) (Murakami et al, 2004) (Kuroda et al, 2001). Clipping is often difficult due to complicated and fragile moyamoya-like vessels in the operative field, moreover, surgical treatment in posterior cranial fossa are relatively complicated and dangerous (Kuroda et al, 2001). Detachable coils are now widely used to treat ICA which is a minimally invasive therapeutic approach, avoiding patients some of the hazards associated with craniotomy and surgical clipping. Therefore, endovascular treatment may be particularly suitable for moyamoya related ICA (Burns et al, 2009).

However, peripheral artery aneurysms commonly distribute in deep brain and the parent arteries are diffusely narrowed. For this reason, microcatheter is usually unable to reach aneurismal lumen; this means that the aneurysm could not be embolized with coils. Under the circumstance, liquid embolic agent embolization may be a worthy choice.

Systematic review of the literature was performed via PubMed search (key words: Moyamoya disease, aneurysm, glue), with careful manual review of the references from relevant articles. As expected articles retrieved represented only six intracranial aneurysms of six patients with Moyamoya disease treated by using liquid embolic agent embolization (Weigele et al, 2002) (Kim et al, 2009) (Murakami et al, 2004) (Yu et al, 2010). The above aneurysms were all peripheral artery aneurysms. The embolic materials were NBCA or glubran 2 which belong to adhesive glue. NBCA (N-butyl-Cyanoacrylate) is a monomer acrylic glue which polymerizes on contact with blood, and subsequently causes a permanent occlusion. Glubran 2 is an acrylic glue authorized for use in surgical and endovascular procedures. The glue needs to be mixed with lipiodol before use to enable its fluoroscopic visualization. The co-monomer of Glubran 2 is comprised of a monomer of NBCA and a monomer of MS (owned by GEM). MS allows the monomer of NBCA to polymerize with a smaller exothermic reaction (45 °C) and a slightly longer polymerization time. Compared with the monomer NBCA, Glubran 2 is associated with a lower risk of adherence of the catheter to the tissue, allowing a greater ease of use (Leonardi et al, 2002) (Yakes et al, 1997). Therefore, the aneurysm can be embolized with glue (glubran 2) at appropriate concentration. Before glue embolization, deep penetration of microcatheter is needed. In the second case, we not only accessed the microcatheter near the neck of aneurysm, but also injected low concentration of glue to make the glue to reach the aneurysm avoiding occlusion of the parent vessel only. Maekawa et al reported a ruptured ICA on lateral posterior choroidal artery associated with moyamoya disease embolized with NBCA, but the patient emerged a large hemispheric infarct that extended well beyond the typical LPChA territory, the authors found that the reason of complication was the parent artery provided collateral hemispheric blood...
flow (Maekawa et al, 1999). Weigele et al using NBCA embolized two aneurysms on LPChA of two patients without moyamoya disease, both patients got optimal outcome (Weigele et al, 2002).

Fig 1-A: CT scan shows subarachnoid hemorrhage in suprasellar cistern and cisterna ambiens, 1-B, C, D: Right vertebral angiograms shows a 3-mm saccular aneurysm at left P2 segment of posterior cerebral artery (1-B: Frontal view, 1-C: Lateral view, 1-D: 3D reconstruction view); 1-E, F: Right vertebral angiogram shows occlusion of aneurysm with coils (1-E: Frontal view, 1-F: Lateral view), 1-G, H: carotid angiograms shows obstruction at the terminal portion of internal carotid artery and moyamoya vessels. (Arrow point to aneurysm).

Fig 2-A: CT scan shows right intraventricular hemorrhage; 2-B, C: Left vertebral angiograms shows a 3-mm aneurysm arising from the distal portion of right lateral posterior choroidal artery (1-B: Lateral view, 1-C: Frontal view); 2-D: Superselective angiogram shows a 3-mm aneurysm, 2-E: Left vertebral angiograms shows complete obliteration of the aneurysm. (Arrow point to aneurysm).

We do not recommend onyx embolization for such aneurysm, which is another kind of liquid embolic agent. In comparison with NBCA and glue 2, the diffusion ability of onyx is relatively poor. Onyx may only occlude the parent vessel and could not embolize the aneurysm when the tip of microcatheter could not be placed in the lumen and was in the parent vessel.

**Conclusion**

The endovascular embolization of intracranial aneurysm associated with moyamoya disease is a reasonable and effective treatment. If peripheral artery aneurysms associated with moyamoya disease can not be embolized with detachable coils, “glue” embolization may be an alternative method.

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**References**


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