Endovascular GDC Treatment of an Idiopathic Carotid-Cavernous Fistula Caused by Aneurysmal Rupture of the Intra-Cavernous Carotid Artery

Case Report

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Summary

Aneurysmal rupture of the intra-cavernous carotid artery may cause idiopathic carotid-cavernous fistula (CCF), and the treatment choice for occluding shunting fistula in this type of CCF is an endovascular approach using detachable balloons. However, little has been reported on treating such lesions with the intra-aneurysmal embolization using Guglielmi detachable coils (GDCs). To our knowledge, ours is the first reported case of successful treatment by selective intra-fistula and intra-aneurysmal embolization with GDCs. A 74-year-old woman exhibited proptosis and chemosis of her left eye over a period of one month. Symptoms of double vision in conversion and pulsatile murmur in her left eye were also noted. Angiography revealed an intra-cavernous aneurysm of the left internal carotid artery (ICA) with a shunting fistula, which drained into the dilated cavernous sinus, superior orbital vein (SOV), superior petrosal sinus, inferior petrosal sinus, and pterygoid plexus. We thought the fistula would occlude by intra-aneurysmal embolization, but we had no confidence of tight packing of the aneurysm since the aneurysmal neck was relatively wide. So, we embolized the venous side of the shunting fistula and then the dome of the aneurysm with GDCs. Immediately after the operation, her symptoms and signs were ameliorated, and com-

plete occlusion of the CCF was observed on long-term follow-up. We suggest selective intra-fistula and intra-aneurysmal embolization with GDCs as an alternative method of treatment of idiopathic CCF originating from aneurysmal rupture of the intra-cavernous carotid artery.

Introduction

The most common treatment of direct CCF is currently the endovascular approach. Trans-arterial detachable balloon occlusion of the shunting fistula has been recommended in cases of direct CCF^{4,5}; however, in some instances endovascular balloons may be inappropriate, particularly when the shunting point not exist along ICA, or the shunting fistula or cavernous sinus is too small or too large, or for cases of recurrence due to deflation of a previously-placed balloon. Our case also had the problem that there is an aneurysm between ICA and the fistula. As an alternative, we used selective intra-aneurysmal embolization with GDCs on both the venous side of the shunting fistula and the dome of the aneurysm. Since the treatment procedure used in this patient is very effective and has never been reported in the literature, we introduce the clinical manifestations of our patient, method of treatment, and her outcome and review the literature.

Figure 1 Photographs showing the patient's left eye. Left: before the embolization. Significant chemosis is seen. Right: post-operation. Chemosis has disappeared.

Case Report

A 74-year-old woman developed proptosis and chemosis of her left eye over a period of one month without past history of head or neck trauma, except for aortic valve replacement five years previously due to non-rheumatic aortic valve stenosis. The patient exhibited no external stigmata or other signs of connective tissue disorders. Family history was also not contributory. She had noticed double vision in conversion and pulsatile tinnitus in her left eye over a period of one month. As the symptoms gradually worsened, she consulted a local ophthalmologist, and was soon transferred to our department for further treatment. On admission, she had chemosis and dilatation of retinal veins (figure 1 left), visual impairment (Visual acuity: lt. 0.3; rt. 1.0), elevated intraorbital pres-

sure (lt. 25 mmHg; rt. 16 mm Hg), proptosis (lt. 14 mm; rt. 10.5 mm), double vision in conversion, and a bruit (vascular sound) that could be auscultated with a stethoscope in the left eye. Angiography was performed on May 28, 2004, and the left internal carotid angiograms showed an intra-cavernous aneurysm (4 mm in diameter) with a comparatively broad neck, which drained into the dilated cavernous sinus, superior orbital vein (SOV), superior petrosal sinus, inferior petrosal sinus, and pterygoid plexus. The final diagnosis of idiopathic left CCF (Barrow Type A) responsible for the rupture of an intra-cavernous aneurysm was then confirmed (figure 2).

Enhanced CT performed on May 31 also revealed marked left proptosis and dilatation of the left SOV.

We initially planned selective coil emboliza-

Figure 2 The left CAG before embolization. The ruptured intra cavernous aneurysm, cavernous sinus and dilated superior orbital vein (SOV), superior petrosal sinus, inferior petrosal sinus and superior pterygoid plexus drainages have appeared in arterial phase.

Figure 3 The left CAG after embolization. The CCF was completely obliterated and the left internal carotid artery is patent.

tion of the intra-cavernous carotid arterial aneurysm with GDCs. We also considered an alternative method to guide a non-detachable balloon into the lumen of the fistula, if the fistula is wide enough for a suitable balloon to be able to pass through. On June 9, 2004, a 7 Fr. guiding catheter was inserted under the right femoral artery approach. We introduced a non-detachable balloon (navi-balloon 1.35, Kaueka, Tokyo, Japan) in an attempt to access the fistula, but failed. We then switched to the selective coil embolization of the intra-cavernous aneurysm. We introduced a guide wire to the aneurysm, with the wire guided carefully to the venous side of the fistula orifice near the cavernous sinus.

After confirming the tip of the micro-guide wire exiting the cavernous sinus, we applied a micro-catheter to the point of rupture of the aneurysm and began to detach one coil from the venous side of the fistula orifice (shunting point), and then detached two other coils to the dome of the aneurysm. Since this aneurysm had a relatively broad neck, we also use the balloon-assisted technique through the GDC embolization procedure.

The procedure was carried out with use of a total of three coils, and a total embolization ratio of 26.43% of the aneurysm was obtained. Post-operative angiography revealed no opacity of the cavernous sinus, nor of the SOV, superior petrosal sinus, inferior petrosal sinus, or pterygoid plexus (figure 3). The audible pulsatile sound in the left eye disappeared completely. The impairment of visual acuity, proptosis, and chemosis were also resolved gradually. On June 21, she had a visual acuity of 1.0 in the left, and 1.2 in the right, and her intraocular pressure and degree of exophthalmos (lt. 11 mm rt. 11 mm) were normal

(figure 1 right). On June 23, she was discharged from our department, and follow-up angiography revealed no recurrence of CCF with complete obliteration of the aneurysm.

Discussion

Several clinical classifications of carotid-cavernous fistula (CCF) have been reported, depending on variation in anatomy, etiology, and pathology. Of the various classifications, one commonly used one divides lesions into traumatic and idiopathic (spontaneous) fistulas. The latter are responsible for rupture of intra-cavernous aneurysms, and are associated with connective tissue disorders (Ehlers-Danlos syndrome), and other systemic vascular diseases¹. In the literature, idiopathic CCF has been further divided into direct and indirect types from the perspective of endovascular treatment. Tearing of the intimal layer of the internal carotid artery is the main cause of direct CCF, and intra-cavernous aneurysmal formation is another cause of it. In most cases, symptoms of direct idiopathic CCF are progressive. Because of its high-flow shunting character, spontaneous remission of the fistula is extremely rare, unlike the case for indirect CCF. If patients with direct CCF have visual impairment or cranial nerve dysfunction, treatment should be performed immediately, or irreversible injury may result².

Trans-arterial use of detachable balloon catheters has revolutionized the treatment of direct idiopathic CCF^{6,7}. However in some exceptional conditions^{8,9}, other endovascular treatments using combined detachable balloon occlusion and GDC embolization, trans-arterial GDC embolization, or trans-venous emboli-

zation are alternatives. The exceptional conditions can be summarized as follows:

1. The fistula orifice is too small to allow balloon entry.
2. The venous compartment is too small to allow balloon inflation, or sharp objects (such as bone fracture fragments or foreign objects) may puncture the balloon during inflation.
3. The shunting point not exist along ICA (eg. rupture of aneurysm)
4. Patients have had prior balloon embolization with subtotal occlusion.
5. The presence of embolic material (balloons) partially blocks the fistula orifice. Mass effect compressing the cavernous sinus and causing occlusion of the internal artery or paralysis of the cranial nerves has also been reported².

Since our patient had a relatively small aneurysm with a well-demarcated shunting fistula, we chose selective intra-aneurysmal GDC embolization instead of detachable balloon method. We first detached one coil to the venous side of the fistula orifice, and then used two other coils in the dome of the aneurysm, which prevented the occurrence of mass effect with possible compression of the cavernous sinus and the internal carotid. Since the shunting fistula was completely occluded by the first coil from the venous side, migration of the coil to the cavernous sinus did

not occur, with consequent excellent results. Unlike the former reported two cases^{2,3}, in which only intra-aneurysmal embolization with GDCs was performed, the shunting fistula in our case was completely obliterated by the first coil and intra-aneurysmal embolization was also successfully performed, which prevented new development of dangerous drainage or aneurysmal enlargement.

Conclusions

We experienced a case of idiopathic CCF responsible for rupture of an intra-cavernous aneurysm. Trans-arterial selective GDC embolization of the shunting fistula and the aneurysm was chosen and yielded complete occlusion of the CCF with preservation of the ICA. The patient's symptoms and signs disappeared completely. We suggest that if diagnostic angiography reveals the shape of the aneurysm permits coil embolization with an excessively narrow fistula lumen, selective GDC embolization of the aneurysm is one of the treatment choice.

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