CASE REPORT

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Spontaneous Regression of the Pseudoaneurysm Developed after Balloon Occlusion of the Direct Carotid-cavernous Fistula

Chae Heuck Lee, M.D.,[1] Myoung Soo Kim, M.D.,[1] Ghi Jai Lee, M.D.[2]

Departments of Neurosurgery,[1] Neuroradiology,[2] Seoul Paik Hospital, Inje University College of Medicine, Seoul, Korea

Direct carotid-cavernous fistula (CCF) is a common post-traumatic disease. However, pseudoaneurysm formation after balloon occlusion is a rare complication. The author present such a case with review of the literature. A 26-year-old man involved in a motor vehicle accident as a driver. Only mild conjunctival injection and minimal exophthalmos on the right eye were noted after trauma. However, angiography revealed a direct CCF and dissection of the proximal intracranial internal carotid artery (ICA). After first balloon occlusion of the CCF, the patient redeveloped fistula due to early deflation of the balloon. After the second balloon occlusion, pseudoaneurysm and diplopia were developed with the change of balloon position and shape. However, visual symptom spontaneously resolved and pseudoaneurysm was also decreased within 6 months after balloon occlusion.

KEY WORDS : Carotid-cavernous fistula · Pseudoaneurysm · Balloon occlusion.

Introduction

Direct carotid-cavernous fistula (CCF), an immediate shunting of blood from the internal carotid artery (ICA) to the cavernous sinus is usually post-traumatic, caused by laceration of the ICA or rupture of its intracavernous dural branches. Traumatic fistulas usually occur in young men.

Endovascular balloon occlusion is accepted as the treatment of choice for traumatic CCF, for complete occlusion of fistula and maintenance of patency of carotid artery. We report an unusual complication of balloon occlusion in a direct CCF following a successful embolization of the fistula.

Case Report

This 26-year-old man was involved in a high speed motor vehicle accident and suffered a mild head injury. He was drowsy but without any neurological deficits. Initial brain CT scan showed small contusion on the right frontal base without skull fracture. There was mild conjunctival injection and minimal exophthalmos on the right. Ten days later, facial bone CT scan revealed marked dilated right superior ophthalmic vein and enlargement of right cavernous sinus, consistent with a CCF. Cerebral angiography revealed a right sided CCF, draining anteriorly by superior and inferior ophthalmic vein, posteriorly by inferior petrosal sinus, superiorly by superficial sylvian vein and basal vein of Rosenthal, inferiorly by pterygoid venous plexus. The filling of the tributaries of the right ICA was poor (Fig. 1).

Operation

We performed transarterial embolization of the fistula on the right cavernous sinus by detachable balloon. After introducing a No. 7.0 French guiding catheter into the right cervical ICA, a microcatheter with a tip mounted latex balloon (GVB9, Ingenor Medical System, Paris, France) selected the right cavernous sinus and progressively inflated it until the fistula from the cavernous ICA to the cavernous sinus was not seen. Postembolization angiography revealed a minimal fistula on same site and irregular contour and narrowing of the lumen at the distal ICA above the ophthalmic artery (Fig. 2).

Two days after the embolization, skull x-ray didn't show

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• Address for reprints : Chae Heuck Lee, M.D., Department of Neurosurgery, Seoul Paik Hospital, Inje University College of Medicine, 85 Jeo-dong 2-ga, Jung-gu, Seoul 100-032, Korea Tel : +82-2-2270-0029, Fax : +82-2-2270-0573, E-mail : chleens@ korea.com

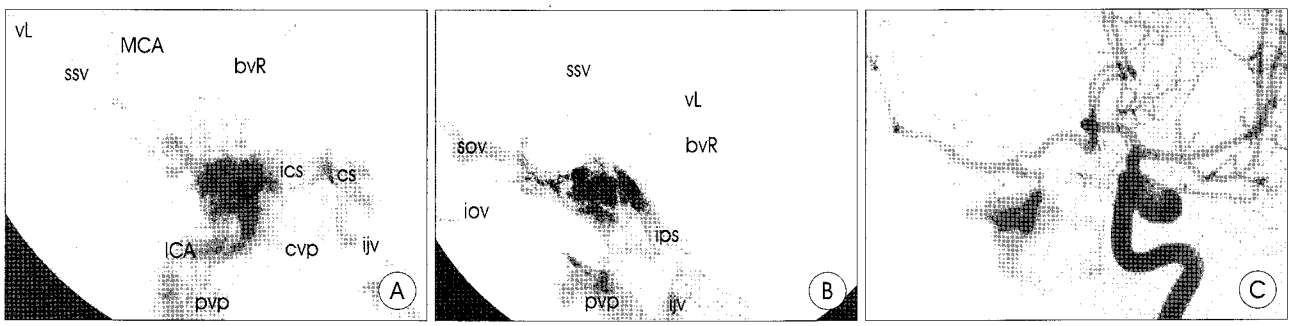

Fig. 1. Selective right internal carotid angiography; A : anteroposterior and B : lateral views revealing a right side carotid-cavernous fistula with filling of superior and inferior ophthalmic vein, inferior petrosal sinus, superficial sylvian vein, basal vein of Rosenthal, and pterygoid venous plexus. The poor filling of the tributaries of the right internal carotid artery and middle cerebral artery was noted. C : Selective left internal carotid angiography with right cervical carotid compression showing retrograde filling of the right cavernous sinus. bvR : basal vein of Rosenthal, cs : cavernous sinus, cvp : clival venous plexus, ICA : internal carotid artery, ics : intercavernous sinus, ijv : internal jugular vein, iov : inferior ophthalmic vein, ips : inferior petrosal sinus, MCA : middle cerebral artery, pvp: pterygoid venous plexus, sov : superior ophthalmic vein, ssv : superficial sylvian vein, vL : vein of Labbe.

radiopaque balloon and the patient complained of bruit. Repeated angiography showed a recurrent fistula (Fig. 3A). A second embolization was then performed with introduction of microcatheter with a tip mounted another detachable latex balloon (GVB12) to the right cavernous ICA and occluded the fistula (Fig. 3B, C). The following day, the patient complained diplopia. Neurological examination showed 6th cranial nerve palsy. We initiated a 7-day course of steroid treatment and a 6-month course of anticoagulation therapy (clopidogrel and aspirin).

Postoperative course

During the next 3 weeks, bruit was disappeared and diplopia was slightly improved. Follow-up angiography (Fig. 4A) obtained at 3 weeks demonstrated pseudoaneurysm at the fistula site of the cavernous sinus. During 6 months, 6th cranial nerve palsy was completely resolved and size of the pseudoaneurysm decreased (Fig. 4B).

Discussion

The key to success of treatment of any CCF is a full understanding of the intracranial blood flow. Bilateral carotid and vertebral angiography with manual cross-compression study is mandatory. The exact size and site of the fistula should be clarified before attempting its treatment. A low- or high-flow fistula should also be assessed subjectively. Additionally, assessment of the clinical or electroencephalographic tolerance to manual carotid compression may be required[7].

Spontaneous closure of a traumatic CCF has been reported rare[2,4]. Occlusion also reported after an angiography or intermittent carotid compression, possibly from a reduction of blood flow in a rare, low-flow fistula sufficient to promote thrombosis[6,7]. Unfortunately, most traumatic CCF show high-flow shunt. The speed of the shunting is the most important factor determining whether a fistula will need aggressive treatment or close observation to thrombose spontaneously.

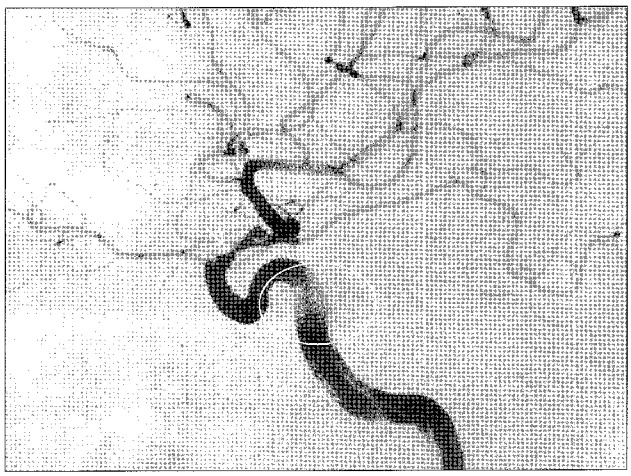

Fig. 2. Angiography after latex balloon (within white circle) embolization closed the fistula with preservation of cerebral blood flow but demonstrating the irregular narrowing of the lumen at the distal internal carotid artery above the ophthalmic artery (arrow).

A variety of transarterial or transvenous approaches and embolic materials are available in the treatment of CCFs, at present. The standard treatment of direct CCF is to occlude the fistula completely while maintaining a patent carotid flow. Despite the development of transvenous approaches and improved techniques in cranial base surgery, transarterial detachable balloons remain the best initial therapy for direct CCFs. However, there is potential risk of ICA occlusion for several reasons[5]. When there is large tear or a nearly complete transection of the ICA, preservation of the ICA is nearly impossible. Sometimes balloon may protrude into the lumen of the ICA and develop a significant stenosis or thromboembolism. Balloon overinflation or early deflation and even rupture may cause recurrence of the CCFs or occlusion of the ICA or potentially resulting in fatal subarachnoid

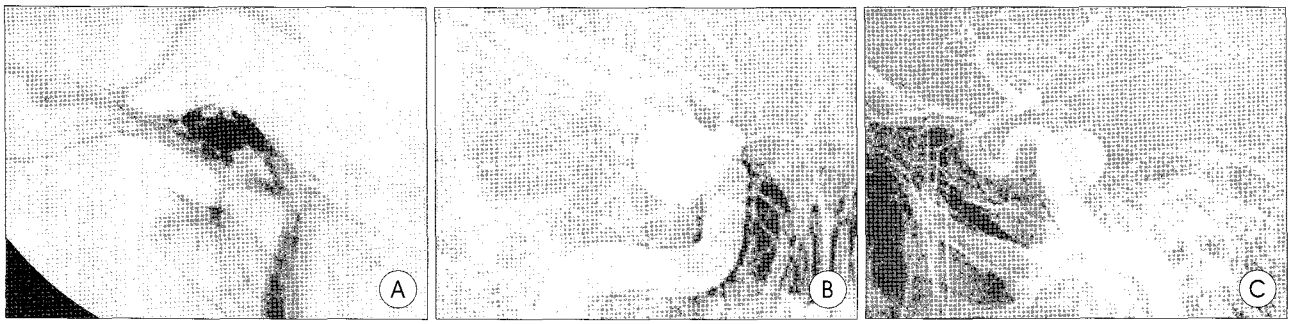

Fig. 3. Repeated right carotid angiography lateral view (A) showing a recurrent fistula after early deflation of the balloon. After second successful obliteration of the fistula with a latex detachable balloon, selective right internal carotid angiography revealing two balloon makers and preservation of the blood flows (B : anteroposterior view and C : lateral view).

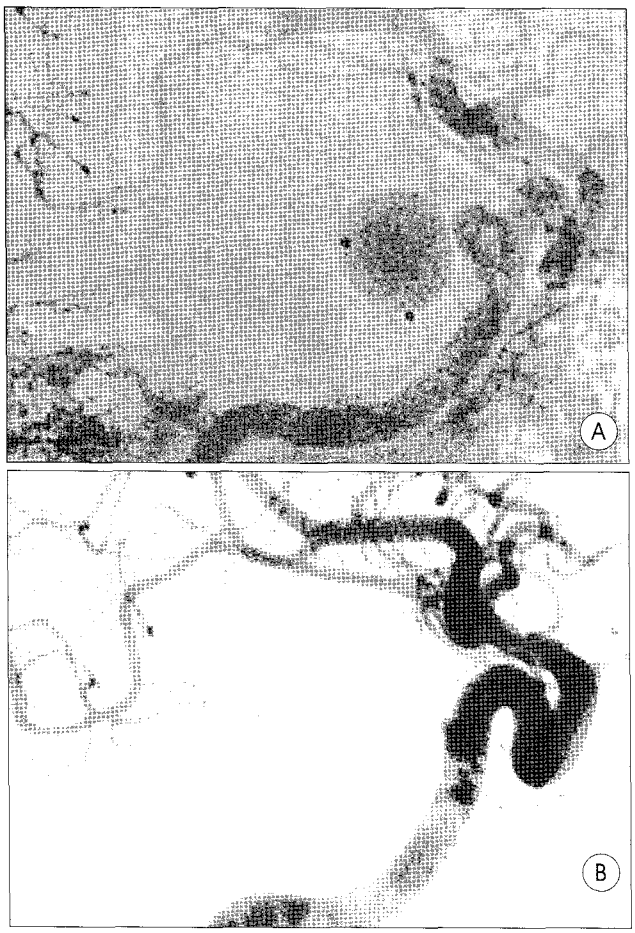

Fig. 4. Three weeks after second balloon occlusion, follow-up angiography (A) showing a slightly deflated balloon displaced from the cavernous sinus. Newly developed pseudoaneurysm is shown on the cavernous portion of the internal carotid artery. Six months later, pseudoaneurysm is decreased on the same projection angiography (B).

hemorrhage. After the balloon occlusion, it should be kept for about 48 hours due to the thrombosis of the cavernous sinus. After detaching the inflated balloon, it will shrink progressively and deflate totally after about a month. Premature deflation or displacement of the balloon may cause development of a pseudoaneurysm that may produce cranial nerve palsy or retro-orbital pain[5].

Pseudoaneurysms are remnants of the wall defect of the ICA at the site of the previous fistula that form after the detachable balloon deflates. It's incidence is 2.8 to 18%[3]. It has the potential to enlarge that may cause mass effect on surrounding cavernous structure, and rupture which may lead to a recurrent CCF, or develop a thrombus that can obliterate the distal cerebral arteries. However, it may remain asymptomatic or decrease in size[5].

The complications of occlusion with detachable balloon are fistulae recurrence, pseudoaneurysm formation, foreign body reaction, cranial nerve paralysis, and cerebral ischemia[1,5]. In our case, the first trial of balloon occlusion resulted in spontaneous deflation of the balloon and recurrence of the CCF. The second trial balloon was also progressively changed its position and shape. As a result, asymptomatic pseudoaneurysm was formed within the cavernous sinus. Post-occlusion 6th cranial nerve palsy might be due to compression of the overinflated detachable balloon or deflated balloon material. After deflation of the balloon, deflated one still might have reduced the flow within the fistula, changing it into a low-flow one and promoting thrombosis. Thrombosis also may have decreased the volume and pressure of the cavernous sinus and subsequently improved the cranial nerve symptoms.

Conclusion

We experienced an unusual complication of detachable balloon occlusion in a direct CCF and spontaneous regression of the pseudoaneurysm. Transarterial balloon embolization technique is a standard method of direct CCFs while preserving a patent parent ICA. Even after a successful occlusion of the fistula, there is a possibility of development of unusual complications such as fistula recurrence, pseudoaneurysm and cranial nerve palsy. Therefore, daily skull X-ray for several days post-operatively and a regular follow-up angiography is recommended, even in asymptomatic patients.

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