Original Article

Clinical Outcomes of Endovascular Treatment for Carotid Cavernous Sinus Fistula at a Tertiary Hospital

Suntaree Thitiwichienlert*, Chonwarat Phattarapongdilok*, Dilok Tantongtip**

Abstract

Background: A carotid-cavernous sinus fistula (CCF) is an abnormal arteriovenous communication between the cavernous sinus and the internal carotid artery (ICA) and or external carotid artery (ECA). The goal of endovascular treatment is the closure of the fistula with preservation of carotid artery patency.

Purpose: To report clinical outcomes using embolic material for the treatment of patients with CCF.

Design: Retrospective study

Materials and methods: The authors retrospectively reviewed the medical records of 25 patients with CCF that underwent the endovascular treatment and follow up from January 2015 to December 2018.

Results: Based on the angiographic characteristics following the Barrow classification, there were 15 patients of the type A CCF, 8 patients of indirect CCF and 2 patients of the combine type CCF. The embolic materials included 48% of detachable balloon, 32% of glue, 8% of coils, and 12% of combined materials. There were 84% of complete embolization, 16% of incomplete embolization with residual fistula. Twenty-three patients (92%) of fistula were totally occluded with the preservation of the carotid artery. There were 2 patients developed stroke due to ICA insufficiency.

Conclusion: Endovascular treatment to embolize the different types of CCFs had a good successful rate with the majority of patients had improvement of the visual function.

Keywords: Carotid cavernous sinus fistula, Cerebral angiography, Endovascular treatment

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Background

A carotid-cavernous sinus fistula (CCF) is an abnormal arteriovenous communication between the cavernous sinus and the internal carotid artery (ICA) and/or external carotid artery (ECA). CCF can be classified according to these criteria: 1) etiology into traumatic or spontaneous CCF; 2) velocity of blood flow into high flow or low flow CCF; and 3) anatomy into direct or indirect CCF.[1] In 1985, Barrow et al. classified CCF into 4 angiographic categories. Type A fistulas are formed by a traumatic tear in the cavernous portion of the ICA resulting in a high blood flow direct shunts between the cavernous sinus and ICA. Types B, C, and D fistulas are formed by a spontaneous connection in meningeal branches of the carotid artery resulting in a low blood flow indirect or dural shunts between the cavernous sinus and meningeal branches of the ICA, ECA and both, respectively.[2, 3]

The clinical features of direct CCF are more severe symptoms and signs than indirect CCF. Direct CCF rarely closes spontaneously and requires treatment if there is visual deterioration, obstructive diplopia, intolerable bruit or headache, or significant proptosis. Previous case series in the literature reported that indirect CCF had incidence of spontaneous resolution from 20% to 60%.[4] Most cases of indirect CCF received initial conservative treatment, but the patient still had to be closely monitored with visual acuity, intraocular pressure (IOP), proptosis measurement, gonioscopy, pupillary function, dilated fundus examination and visual fields. Endovascular treatment is proposed when the patient has progressive visual loss or signs of posterior cortical venous drainage.[5, 6] In the present study, the authors reported the clinical features and our results with endovascular treatment of CCF using transarterial, transvenous or combine an approach for embolization.

Materials and methods

The study was approved by the Medical Ethics Committee of Thammasat University (MTU-EC-OP-1-279/61), Pathum Thani, Thailand, and was conducted in accordance with the tenets of the Declaration of Helsinki. The authors retrospectively reviewed the medical records of patients with CCF that underwent endovascular treatment in the Department of Ophthalmology and Department of Surgery, Thammasat Hospital, Thammasat University, Thailand, from January 2015 to December 2018.

Demographics and clinical characteristics

A total of 39 eyes in 36 patients were enrolled. There were three patients who did not need to embolize the fistula. The authors retrospectively reviewed the data of 36 eyes in 33 patients who underwent embolization of a CCF from 2015 to 2018 at our center. Eleven patients with missing data were excluded. Twenty-five patients (28 eyes) were included in the study. They had a mean age of 44.7 ± 18.5 years (range 17 to 75 years). Fifteen patients were male and ten were female. The most common etiology of direct CCF was head trauma. There were 16 patients (64%) of traumatic direct CCF and 2 patients (8%) of spontaneous direct CCF. There were 7 patients (28%) of spontaneous indirect CCF.

The lateralization of CCF depends on the venous drainage route, including the inter-cavernous communication, the cortical venous drainage, and the thrombosis on one or both sides.[7, 8] Unilateral CCF may present with bilateral or even contralateral ocular manifestations. There were 3 patients of bilateral direct CCF, 12 patients of unilateral direct CCF and 10 patients of unilateral indirect CCF. Two patients had unilateral indirect fistulas with bilateral ocular manifestations from communications between the cavernous sinuses. Figure 1 shows an example case

of a post-traumatic unilateral CCF with contralateral symptoms in the fellow eye. Cerebral angiography showed the abnormal shunting from the meningo-hypophyseal trunk of the left cavernous segment ICA to the left cavernous sinus and drain to the right cavernous sinus via the intercavernous sinus with reflux to the right SOV (Figure 1). Cerebral angiography showed the complete occlusion of the fistula after the glue embolization of dural CCF (Figure 2).

Based on the angiography characteristics of the CCF, there were 15 patients (60%) of type A CCF, one patient (4%) of type C CCF, 7 patients (28%) of type D CCF, and 2 patients (8%) of combine type CCF.

The ocular symptoms would often be combined with other signs to reach a diagnosis. These included cranial bruit, proptosis, conjunctival chemosis, corkscrew-type blood vessels, ophthalmoplegia, glaucoma, and visual loss. A cranial bruit may result from the turbulent flow of blood in an artery.[9] Ophthalmoplegia may result from ocular motor cranial nerves dysfunction or extraocular muscles congestion. Glaucoma may result from increased episcleral venous pressure, orbital congestion, choroidal effusion, or neovascular glaucoma.[10] Visual loss may result from traumatic optic neuropathy (TON), exposure keratopathy, venous stasis retinopathy, or ocular hypoperfusion.[11]

In the present study, there were 20 patients (80%) who had bruit, 15 patients (60%) with proptosis, 21 patients (84%) that had corkscrew-type blood vessels, 21 patients (84%) with ophthalmoplegia, 13 patients (52%) with glaucoma, 9 patients (36%) who had venous stasis retinopathy, and 9 patients (36%) with TON. There was one patient (4%) that had central retinal vein occlusion (CRVO), one patient (4%) with central retinal artery occlusion (CRAO) from a steal phenomenon in the cerebral vessels, and one patient (4%) who had a corneal ulceration as a result of significant proptosis and exposure keratopathy. This patient had dense central corneal scarring and ready to reserve the corneal donation from the Thai Red Cross Eye Bank. Though CCF was not life-threatening, there were several reports of fatal non-ocular manifestations such as epistaxis, subarachnoid hemorrhage, or intracerebral hemorrhage from rupture of CCF. A posterior-draining CCF can cause cortical venous reflux and intracerebral hemorrhage as a result of venous hypertension.[12] Fortunately, there was no patient with cortical venous reflux had intracerebral hemorrhage.

Endovascular procedures

All patients underwent brain computerized tomography (CT)/CT angiography (CTA) or magnetic resonance imaging (MRI)/magnetic resonance angiography (MRA). To confirm clinical manifestations of CCF, cerebral angiography was performed in all patients. All endovascular procedures were performed by a single neurosurgeon. For the transarterial approach, all patients were accessed via a right femoral artery and placed a guiding catheter at a cervical segment of ICA or ECA depend on feeding artery location. Then a microcatheter with a micro guidewire was used to navigate into the feeding artery. Then injected the embolic material which commonly used including detachable balloon (only for direct type CCF), coils or n-BCA (n-butyl cyanoacrylate) to occlude the fistula. For the trans-venous approach which usually used in complex fistula patients with small multiple feeding arteries. The route of the approach depended on venous drainage pattern which could identify from angiography including inferior petrosal sinus (IPS) or superior ophthalmic vein (SOV). For the IPS drainage pattern, all patients were accessed via a right femoral vein and placed the guiding catheter at an internal jugular vein then navigated the microcatheter with the micro guidewire via IPS to cavernous sinus. The most common embolic materials were coils and n-BCA. For SOV drainage pattern, all patients were directly assessed in the SOV via a small incision at the eyelid by placing the guiding catheter at the cavernous sinus and then placing the embolic material in the same way as the trans-IPS.

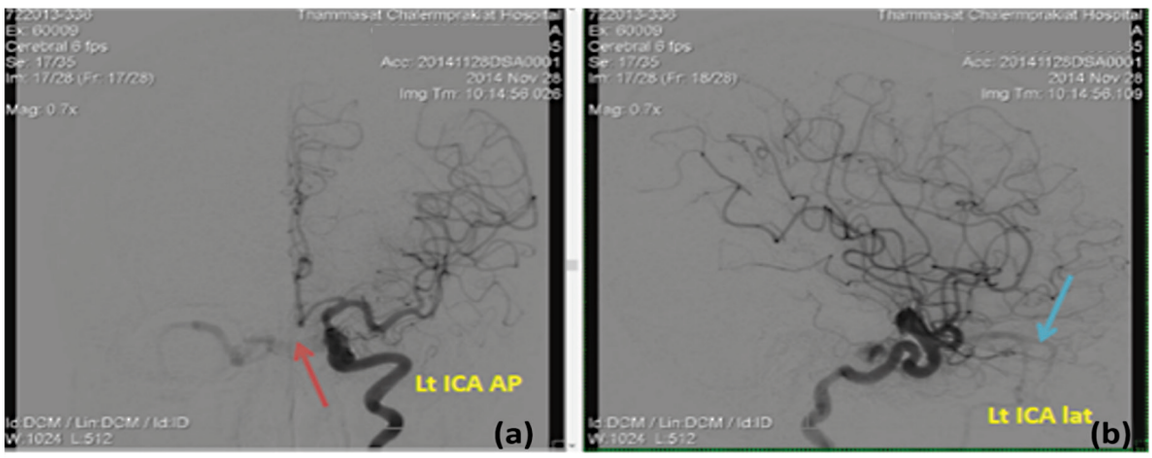

Figure 1 Cerebral angiography (a) showed the abnormal shunting from the meningo-hypophyseal trunk of the left cavernous segment ICA to the left cavernous sinus and (b) showed the shunting drain to the right cavernous sinus via the intercavernous sinus (red arrow) with reflux to the right SOV (blue arrow).

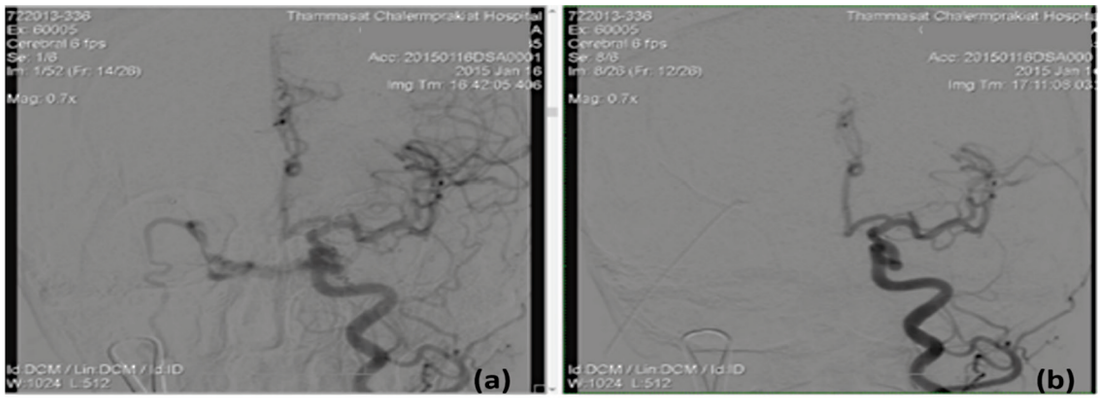

Figure 2 Cerebral angiography (a) showed transarterial glue embolization and (b) showed the complete occlusion of the fistula after the glue embolization of dural CCF.

Embolization of CCF using materials was performed in 12 patients (48%) treated with detachable balloons, 8 patients (32%) treated with glue, 2 patients (8%) treated with coils, one patient (4%) treated with balloons and glue, one patient (4%) treated with balloons and coils, and one patient (4%) treated with glue and coils.

Results

Angiographic results

The embolization procedure was successful in 21 patients (84%) and unsuccessful in 4 patients (16%). One of the residual fistulas could not be occluded due to the fistula being too small to be cannulated. Two patients failed the first endovascular

treatment. The first patient had multiple indirect types D CCF mostly from the left ECA. The second patient had an incidental finding of an unruptured left posterior communicating artery aneurysm. The first patient underwent a craniotomy, extradural anterior clinoidectomy, and transcavernous approach for the CCF disconnection, postoperative cerebral angiography showed complete occlusion of CCF without cortical reflux. The second patient underwent surgical clipping, and the postoperative cerebral angiography also showed a complete occlusion of CCF.

Clinical results

Most patients had clinical improvement after endovascular treatments.These included the disappearance of bruit and corkscrew-type blood vessels in 72% (18 patients), the loss of proptosis in 56% (14 patients), recovery of ophthalmoplegia in 48% (12 patients), recovery of secondary glaucoma in 32% (8 patients), recovery of venous stasis retinopathy in 28% (7 patients), and improvement of visual deterioration in 16% (4 patients). Seven patients had a final acuity worse than 20/200 in at least one eye from TON; 5 patients had a final acuity no light perception (NPL) and 2 patients had a final acuity light projection (PJ).

Surgical complications

The carotid artery was totally patent in 23 patients (92%). Two patients (8%) had a stroke from ICA insufficiency. The first patient had left hemiparesis and alteration of consciousness on the first postoperative day. However, the patient underwent emergency right superficial temporal artery to middle cerebral artery (STA-MCA) bypass, and postoperative cerebral angiography showed complete occlusion of the right direct CCF with good collateral blood flow to the right MCA from the left ICA. The second patient had a history of earlier quadriparesis from bilateral MCA infarction and he had a new onset of left ICA occlusion on the first postoperative day. Fortunately, the patient underwent craniotomy with ICA arteriotomy with balloon removal, and postoperative cerebral angiography showed nearly complete occlusion of bilateral direct CCF. During a follow-up period of 3 months after embolization, one patient had minimal residual fistula at posterior genu of ICA with minimal drainage to the inferior petrosal sinus. However, the patient underwent the second embolization and was successfully treated with coils (Tables 1 and 2).

Table 1 Summary of the data of all reported 25 patients with CCFs.

Case No.	Age (years)	Gender	Etiology	Barrow type	Materials	Patency of ICA	Complication	Closure of CCF
1	50	M	traumatic	A	balloons	no	stroke	complete
2	39	M	traumatic	A	balloons	yes	no	complete
3	37	M	traumatic	A	balloons	yes	no	complete
4	20	M	traumatic	A	balloons	yes	no	complete
5	31	F	traumatic	A	balloons	yes	no	complete
6	18	M	traumatic	A	balloons	yes	no	complete
7	24	M	traumatic	A	balloons	yes	no	complete
8	64	M	traumatic	A	balloons	yes	no	complete
9	57	M	traumatic	A	balloons	yes	stroke	incomplete
10	31	M	traumatic	A	balloons, glue	yes	no	complete
11	27	M	traumatic	A	balloons	yes	no	complete
12	17	M	traumatic	A	balloons	yes	no	complete
13	35	M	traumatic	A	balloons	yes	no	complete
14	26	M	traumatic	A	balloons, coils	yes	recurrence	complete
15	58	F	traumatic	A	coils	yes	no	complete
16	70	F	spontaneous	C	glue	yes	no	complete
17	60	M	spontaneous	D	glue	yes	no	complete
18	75	F	spontaneous	D	glue	yes	no	complete
19	56	F	spontaneous	D	glue	yes	no	complete
20	43	F	spontaneous	D	glue	yes	no	complete
21	32	M	spontaneous	D	glue	yes	no	complete
22	73	M	spontaneous	D	coils	yes	no	complete
23	74	F	spontaneous	D	coils, glue	yes	no	incomplete
24	48	F	spontaneous	combine	glue	yes	no	incomplete
25	53	F	spontaneous	combine	glue	yes	no	incomplete

Table 2 The numbers and percentage of patients' demographic data, etiologies, the Barrow classification, and endovascular treatment.

	Total (N=25)	
	Numbers	Percentage
Gender		
male	15	60
female	10	40
Etiologies of CCF		
traumatic direct CCF	16	64
spontaneous direct CCF	2	8
spontaneous indirect CCF	7	28
Barrow classification of CCFs		
type A	15	60
type B	-	
type C	1	4
type D	7	28
combine type	2	8
Embolic materials		
detachable balloons	12	48
glue	8	32
coils	2	8
detachable balloons and glue	1	4
detachable balloons and coils	1	4
glue and coils	1	4
Closure of CCF		
complete	21	84
incomplete	4	16
Patency of ICA		
yes	23	92
no	2	8

Discussion

The goal of endovascular treatment was the closure of the fistula with the preservation of carotid artery patency.[13, 14] Previous case series reported a success rate between 86%-94% with complication rates of 4%-5.5%. The present study also showed that endovascular treatment had a high success rate. Most patients were successful in the first treatment, whereas others had to undergo a second treatment to achieve success.

A variety of materials can be used to close the fistula. Several techniques can be performed such as embolization with glue, detachable balloons, platinum coils, or combine techniques. In the present study, the authors selected detachable balloons as the first therapeutic cerebral angiography due to the effectiveness to occlude the direct fistula that was relatively high with a low cost compared with the coils. However, the limitations of the detachable balloon including balloon is indicated for only direct type CCF, recurrent during early deflate of balloon and stroke due to early detach of the balloon. A coil is an increasingly used material because it is a permanent embolic that can be accessed from both the transarterial or transvenous approach and able to be navigated through the small fistula. Moreover, the coil could be used in both direct and indirect type CCFs. Therefore, the disadvantages of the coil includes the cost and the amount needed to occlude the fistula due to the large venous drainage chambers. n-BCA is mostly used to occlude the indirect type CCF, as n-BCA can be injected through a small microcatheter which can be used to embolize the feeding small artery. The disadvantages of n-BCA include the risk of stroke due to reflux of the material into parent vessels (which would need the access to the feeding artery), irritation due to chemical reaction and residual fistula which can be occurred because n-BCA can not penetrate to the location of the f istula and leave the small arterial channel to fistula. In 1974, Serbinenko reported a successful method of closing the fistula using a detachable balloon[15]. The deflated balloon is small enough to pass through the fistula into the cavernous sinus and it can be inflated as big as being expected. The flow-guided balloon is attached to the catheter that is advanced up the ipsilateral ICA and into the cavernous sinus. Once the balloon is in proper position in the cavernous sinus, it is inflated and detached, occluding the fistula but leaving the ICA patency. Among 15 patients with direct type CCF, 12 patients were treated with detachable balloons, and three patients were treated with detachable balloons and glue/coil combinations.

Uncommonly, few reports have described the ICA occlusion in patients who underwent balloon occlusion.[16, 17] In the present study, there were two patients who had suffered a stroke from the ICA occlusion following the balloon embolization. These complications were related to the interruption of the blood flow to the cerebral hemisphere, while the collateral vessels were insufficient to supply the ipsilateral cerebral hemisphere. However, these patients had appropriate treatments and postoperative cerebral angiography showed nearly complete occlusion of CCF.

Once a fistula is successfully closed, most of the ocular manifestations can be resolved or at least improved. Postoperative recovery depends on the severity of the fistulas at the time of treatment. Most patients recovered themselves, whereas others experienced persisting residual fistulas. Bruit disappeared immediately, while the corkscrew-type blood vessels, venous stasis retinopathy, ophthalmoplegia, and intraocular pressure slowly dissipated during a period of weeks to months. Patients with TON or retinal dysfunction had poor visual prognosis.

A limitation of the study was the retrospective method. There were a limited number of patients. Some patients were excluded due to incomplete medical record. Another limitation was the inability to assess the exact outcome of treatment, because

the present study was conducted in a tertiary center where most of the cases were more severe and complicated. In conclusion, endovascular treatment to embolize the different types of CCF was quite successful with the majority of patients having improved visual functions.

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Potential conflicts of interest. All authors report no conflicts of interest relevant to this article. The authors wish to thank the Department of Ophthalmology, Faculty of Medicine, Thammasat University for research funding and provision of facilities.

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บทคัดย่อ

ลักษณะทางคลินิกและการรักษาผ่านหลอดเลือดของผู้ป่วยภาวะหลอดเลือดแดงรั่วเชื่อมต่อแอ่งหลอดเลือดดำที่บริเวณฐานสมอง ในโรงพยาบาลตติยภูมิ

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บทนำ: ภาวะหลอดเลือดแดงรั่วเชื่อมต่อแอ่งหลอดเลือดดำที่บริเวณฐานสมองคือ ภาวะหลอดเลือดแดง internal carotid artery และหรือ external carotid artery รั่วหรือมีการเชื่อมต่อแอ่งกับหลอดเลือดดำ cavernous sinus ที่บริเวณฐานสมอง วัตถุประสงค์ในการรักษาผ่านหลอดเลือดคือ การปิดรอยรั่วของหลอดเลือดโดยที่ไม่ทำให้เกิดการอุดตันของหลอดเลือด carotid artery

วัตถุประสงค์: เพื่อศึกษาลักษณะทางคลินิกและผลสำเร็จของการรักษาผ่านหลอดเลือดโดยการใช้วัสดุอุดรอยรั่วของหลอดเลือด

รูปแบบการศึกษา: การศึกษาแบบพรรณนาย้อนหลัง

วัสดุและวิธีการ: ผู้นิพนธ์ทำการศึกษาจากเวชระเบียนย้อนหลังของผู้ป่วย 25 ราย ที่มีภาวะหลอดเลือดแดงรั่วเชื่อมต่อแอ่งหลอดเลือดดำที่บริเวณฐานสมอง ที่ได้รับการรักษาผ่านหลอดเลือด ตั้งแต่เดือน มกราคม พ.ศ.2558 ถึงเดือน ธันวาคม พ.ศ.2561

ผลการศึกษา: จากการแบ่งชนิดของรอยรั่วของหลอดเลือดตามลักษณะของการฉีดสีใน Barrow classification พบว่ามีผู้ป่วย 15 รายเป็นชนิด direct type A, 8 ราย เป็นชนิด indirect type และ 2 ราย เป็น combine type วัสดุที่ใช้อุดรอยรั่วของหลอดเลือด ได้แก่ detachable balloon ร้อยละ 48, glue ร้อยละ 32, coils ร้อยละ 8 และใช้วัสดุอุดมากกว่าหนึ่งอย่าง ร้อยละ 12 มีผู้ป่วยร้อยละ 84 ที่อุดรอยรั่วของหลอดเลือดได้สมบูรณ์ ส่วนผู้ป่วยอีกร้อยละ 16 อุดรอยรั่วได้ไม่สมบูรณ์และยังมีรอยรั่วของหลอดเลือดหลงเหลือ ผู้ป่วย 23 ราย (ร้อยละ 92) ได้รับการรักษาอุดรอยรั่วของหลอดเลือดได้สมบูรณ์โดยที่ไม่เกิดการอุดตันของหลอดเลือด carotid artery มีผู้ป่วย 2 ราย เกิดภาวะสมองขาดเลือดจากการลดลงของหลอดเลือด internal carotid artery

สรุป: การรักษาผ่านหลอดเลือดในผู้ป่วยภาวะหลอดเลือดแดงรั่วเชื่อมต่อแอ่งหลอดเลือดดำที่บริเวณฐานสมองชนิดต่างๆ ได้ผลสำเร็จที่ดี และผู้ป่วยส่วนใหญ่มีการดีขึ้นของ visual function

คำสำคัญ: ภาวะหลอดเลือดแดงรั่วเชื่อมต่อแอ่งหลอดเลือดดำที่บริเวณฐานสมอง, การฉีดสีหลอดเลือดสมอง, การรักษาผ่านหลอดเลือด