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Endovascular Treatment in Traumatic and Spontaneous Carotid Cavernous Fistulas: with Different Embolization Agents and via Various Vascular Routes

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Abstract

Purpose—We evaluated carotid cavernous fistula (CCF) cases which were treated with various vascular routes and different embolization agents. Our aim was to present endovascular treatment procedures, clinical and radiological findings, and to discuss the safety and clinical efficacy of the treatment.

Materials and Methods—The demographic information, presenting symptoms and clinical findings of 25 CCF cases in 23 patients treated with endovascular route were reviewed. The type of fistula, the feeding arteries, the draining veins, and the details of the treatment were documented on the basis of digital subtraction angiography (DSA) images. The efficacy of the treatment was evaluated according to current and follow-up DSA findings with clinical symptoms.

Results—All of which 25 CCF were closed to the fullest extent, one of them closed spontaneously, 20 were closed in one session and 4 in two sessions (100%). 18 of the cases (75%; 18/24) were treated with a venous approach, 5 cases (21%; 5/24) with an arterial approach, and 1 case (4%; 1/24) with a combined arterial-venous approach. Coils were used in 18 cases (75%; 18/24), a covered stent was used in 1 case (4%; 1/24), a detachable balloon was used in 1 case (4%; 1/24), n-Butyl Cyanoacrylate was used in 1 case (4%; 1/24), and combined (2 stent-coil, 1 coil-ethylene vinyl alcohol copolymer) embolization agents were used.

Conclusion—The endovascular treatment of CCF has high success and low complication rates. The significant point of the treatment is achieving complete fistula obliteration in the least possible number of sessions with appropriate embolization agents.

Introduction

Carotid cavernous fistula (CCF) is an abnormal connection between the carotid artery and the cavernous sinus system. CCFs are classified as high or low flow according to the hemodynamic status of the fistula, as traumatic or spontaneous according to the etiology, as direct or indirect according to the arterial supply status on angiography [1–3]. The association between arterial and venous beds results in abnormally increased pressure in the cavernous sinus which causes retrograde and arterialized flow in the veins that drain here, especially in the superior ophthalmic vein (SOV). This mechanism is responsible for the main clinical symptoms of CCF, such as proptosis, chemosis, diplopia, and loss of vision [2]. The primary principle in the treatment is to obliterate the fistula track while maintaining the patency of the arteries [4]. For this purpose, there are many options such as conservative approaches or surgery; however, in line with technological advances, the current treatment of choice in CCF is endovascular treatment [2].

In this study, we aimed to present the clinical and radiological findings, the endovascular treatment procedure, and to discuss the safety and clinical efficacy of CCF cases undergoing endovascular treatment in our department.

Methods

25 CCF cases in 23 patients on whom performed endovascular treatment in our department between 2009 and 2015 retrieved from the database. The clinical findings and radiological images were evaluated retrospectively with the consent of the patients and the ethics approval of the committee. The demographic information of the patients, their comorbid diseases, the presentation symp-

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toms, the type of the fistula, the feeding arteries, the draining veins, and the details of the endovascular treatment were documented. The efficacy of the treatment was evaluated according to the current and follow-up digital subtraction angiography (DSA) findings with clinical symptoms.

The cases were classified according to their arterial supply status on the DSA images with regard to the classification system defined by Barlow *et al*. [5]. Accordingly, the classification was made as direct (type A) and indirect (types B, C, and D). A direct communication between the cavernous sinus and the internal carotid artery (ICA) (high flow and associated with trauma) was regarded as type A fistula, a communication with dural ICA branches as type B fistula, a communication with dural ECA branches as type C fistula, and communication with both ICA and ECA branches was regarded as type D fistula.

Patient demographics and clinical presentation

Among the 23 patients, 7 (30%, 7/23) were male and 16 (70%, 16/23) were female, and the mean age (mean ± SD) was 61 ± 14. 11 patients (48%, 11/23) had hypertension, 5 (22%, 5/23) had a history of facial trauma, 4 (17%, 4/23) had diabetes mellitus, 4 (17%, 4/23) patients were smokers, and 3 (13%, 3/23) had hyperlipidemia. The most common symptoms were proptosis in 18 (78%, 18/23) patients, chemosis in 17 (74%, 17/23), and diplopia in 17 (74%, 17/23). 10 patients had orbital pain (43%, 10/23) and 9 (39%, 9/23) had loss of vision (Table 1).

CCF characteristics

All patients underwent diagnostic DSAs including selective internal and external carotid angiography. The DSAs were evaluated by two neuro-interventional radiologists. The cases had a total of 25 CCFs; 21 were unilateral (12 left, 9 right), and 2 were bilateral. According to the Barlow classification, 5 of the CCFs were type A (20%; 5/25), 4 were type B (16%, 4/25), 3 were type C (12%; 3/25), and 13 were type D (52%; 13/25).

Unlike the type A CCFs that were supplied directly by the ICA, the indirect CCFs (types B, C, and D) were supplied by various branches of the ICA and ECA. Some indirect CCF cases had bilateral supplies. 19 (76%; 19/25) of the cases were supplied by the arteries of the meningohypophyseal trunk (MHT), and 7 (28%; 7/25) were supplied by the arteries of the inferolateral trunk. The ECA branches supplying the CCFs were as follows: the middle meningeal artery in 14 cases (56%, 14/25), the deep temporal artery in 8 (32%; 8/25), the accessory meningeal artery in 5 (20%; 5/25), the artery of foramen rotundum in 5 (20%; 5/15), the ascending pharyngeal artery in 4 (16%; 4/25), and the zygomatico-orbital artery in 1 (4%; 1/25) case.

Table 1. (A) Demographics and clinical presentations for 23 patients of CCF. (B) Morphological characteristics of 25 CCF undergoing multimodality treatment

(A) Demographics and clinical presentations	
Number of patients (*n*)	**23**
Gender	
Men (*n*)	7 (30%)
Women (*n*)	16 (70%)
Age (years), mean ± SD	61 ± 14
Comorbidities	
Hypertension	11(48%)
Fascial trauma	5 (22%)
Diabetes mellitus	4(17%)
Smoking	4 (17%)
Hyperlipidemia	3 (13%)
Clinical presentation	
Propitozis	18 (78%)
Chemosis	17 (74%)
Diplopia	17 (74%)
Orbital pain	10 (43%)
Visula loss	9 (39%)
(B) Morphological characteristics	
Number of CCF (*n*)	**25**
Barrow classification	
A	5 (29%)
B	4 (16%)
C	3 (12%)
D	13 (52%)
Feeding arteries	
Meningo hypophyseal truncus	19 (76%)
Middle meningeal artery	14 (56%)
Deep temporal artery	8 (32%)
Inferolateral truncus	7 (28%)
Accessory meningeal artery	5 (20%)
Artery of foramen rotundum	5 (20%)
Ascending pharyngeal artery	4 (16%)
Zygomatico-orbital artery	1 (4%)
Draining veins	
Superior ophthalmic vein	25 (100%)
Intercavernous sinus	18 (72%)
Inferior petrosal sinus	14 (56%)
Angular vein	11 (44%)
Facial vein	8 (32%)
Cortical veins	7 (28%)
Superior petrosal sinus	5 (20%)
Inferior ophthalmic vein	2 (8%)

CCF, carotid cavernous fistula

The venous drainage of the cases was as follows: all of the cases drained into SOV (100%; 25/25). 18 cases (72%; 18/25) drained into the intercavernous sinus (ICS), 14 (56%; 14/25) into the inferior petrosal sinus (IPS), 11 (44%; 11/25) into the angular vein (AV), 8 (32%; 8/25) into the facial vein, 7 (28%; 7/25) into the cortical veins, 5 (20%; 5/25) into the superior petrosal sinus (SPS), and 2 (8%; 2/25) drained into the inferior ophthalmic vein (Table 1).

Endovascular treatment

All treatments were performed under general anesthesia under biplane angiography unit with 3D rotational

Table 2. Demography, technique, and outcomes of endovascular treatment in 23 patients with 25 CCF

Patients Age (year)/sex	Bar-low type	Seans	Approachs	Emboli-zan agent	Postop DSA 1	Postop clinic out-come	COMPLIC	Follow-up DSA (mo)	Follow-up clinic(mo)
1: 41/F	D	12	IPS-SPSSOV	—COIL	—Cure	—Cure	—No	—	—24
2: 65/M	D	1	IPS	COIL	Cure	Cure	No		10
3: 62/F	D	1	IPS	COIL	Cure	Cure	No		6
4: 80/F	D (L)	1	IPS	COIL	Cure	Cure	No	3	3
4: 80/F	D (R)	1	IPS	COIL	Cure	Cure	No	3	3
5: 61/M	B	1	AV	COIL	Cure	Cure	No		76
6: 55/F	D	1	IPS	COIL	Cure	Cure	No		24
7: 38/M	A	12	ICAICA	Solit + COIL-COIL	CureCure	CureCure	NoNo	2(nuks)	—2
8: 79/F	D	1	IPS	COIL	Cure	6 palsy	No	3	18(6 palsy)
9: 64/F	D	1	SPS	GLU	Cure	Cure	No		3
10: 48/M	A	1	ICA	Cover Stent	Cure	Cure	No		3
11: 70/F	A	1	IPS	COIL	Cure	3 palsy	No	12	61(3 palsy)
12: 74/F	D	1	IPS	COIL	Cure	Cure	No		77
13: 55/F	B	12	IPS_SPSSOV	—COIL	—Cure	—Cure	—No	—	52
14: 27/M	A	1	ICA	Solit+COIL	Partial	Cure	No	2(complet)	12
15: 76/F	D	1	IPS	COIL	Cure	Cure	6 palsy		27(6palsy)
16: 63/F	C	1	IPS	COIL	Cure	Chemozis	No	3	3(chemozis)
17: 61/F	C	1	AV	COIL	Partial	Cure	No	3(complet)	3
18: 81/F	D	1	IPS	COIL	Partial	Cure	No	3(complet)	20
19: 45/F	A	1	ICA	DetachBalon	Cure	Cure	No	28	28
20: 50/M	D	1	IPS	COIL	Cure	Cure	No		3
21: 62/M	B	Spon-tan	—	—	—	—	—	—	4
22: 63/F	C (L)	1.2.	IPSECA	COILONYX	PartialCure	ChemozisCure	NoMicroen-fart	2(partial)	12(complet)
22: 63/F	D (R)	1.	IPS	COIL	Cure	Cure	No		12
23: 79/F	B	1.	MHT	COIL	Cure	Cure	No	2	2

CCF, carotid cavernous fistula; DSA, digital subtraction angiography; F, female; M, male; IPS, inferior petrosal sinus; SPS, superior petrosal sinus; SOV, superior ophthalmic vein; AV, angular vein; ICA, internal carotid artery; ECA, eksternal carotis artery; MHT, meningohypophyseal trunk.

angiography capacity (AXIOM Artis FD Biplane Angiosuite with DynaCT; Siemens Medical Solutions). Selective DSAs were obtained from the ICA and ECA from both sides and the fistulae were revealed. The type of the fistula and the patient's vascular anatomy were established, and the treatment method and embolization agents that would be used were determined.

Endovascular treatment was performed according to the lesion type, the arterial and venous communications, the vascular anatomy, and variations of the case. In one patient admitted to the angiography unit for treatment, it was observed that the fistula had closed spontaneously. Among the remaining 24 treatments, 18 (75%; 18/24) were performed through a venous approach, 5 (21%; 5/24) through an arterial approach, and 1 (4%; 1/24) through a combined arterial and venous approach. The venous routes were as follows: IPS 13, AV 2, SOV 2, and SPS 1. The arterial routes were direct ICA in four cases, MHT in one case, and IPS + ECA in one case undergoing combined treatment.

Different embolization agents were used in the treatment of the cases according to the type of the fistula. Coils were used in 18 (75%; 18/24) cases, a covered stent was used in 1 (4%; 1/24), a detachable balloon was used in 1 (4%; 1/24), n-butyl cyanoacrylate (n-BCA) was used in 1 (4%; 1/24), and combined embolization agents were used in 3 (13%; 3/24) cases. The combined treatment was used with stent + coil in two cases, and coil + ethylene vinyl alcohol copolymer (Onyx) in one case (Table 2).

Results

Angiography results

11 (48%) of the 23 patients underwent DSA follow-ups. The mean follow-up duration was 2–28 months (7 ± 7). Among the 25 CCFs, 1 closed spontaneously, 20 closed in one session, and 4 closed in two sessions (100%). No endovascular procedures were performed on the CCFs that closed spontaneously. In 3 of the 20 cases (cases number 14, 17, and 18) undergoing 1 session, the DSA performed immediately after the procedure revealed low flow partial filling (residue). These cases were monitored and were seen the fistula had closed completely on the follow-up DSAs at the second and the third months, respectively. A second treatment session was not required (Table 2). Case examples are shown in Figures 1 and 2.

Clinical results and complications

Complications developed in two cases early postoperative period after endovascular treatment. No complica-

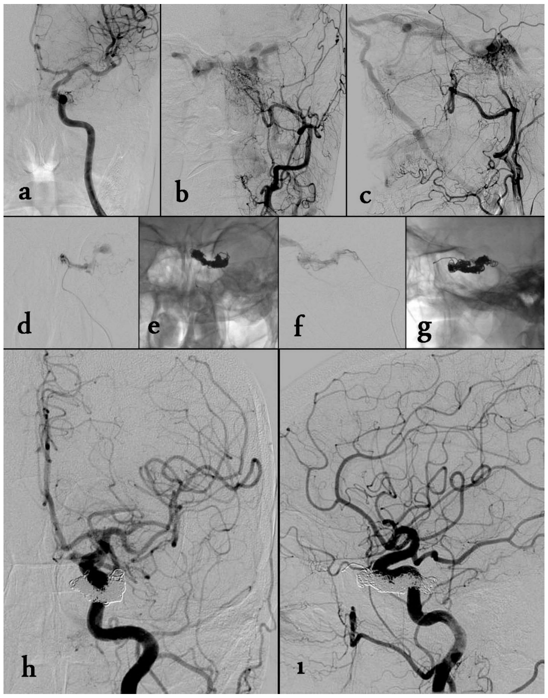

Figure 1. Barlow type C Ccf (case no. 16) which was fed only by the branches of the external carotid artery and drained to the right cavernous sinus through Ics and Sov was detected at the left sided selective internal (a) and external carotid (b) and (c) anjiography of the patient who presented with chemosis, ptosis and orbital pain in the left eye. Total occlusion of the fistula, performed through coil embolization with using of microcatheter from left Ips, was seen in this case (d)–(g) at the postoperative anterioposterior (h) and lateral (i) common carotid artery anjiography.

tions developed in the remaining 21 cases. The two cases that developed complications were as follows (Table 2).

Case 1 (Case number 15): This case underwent coil treatment through the IPS for type D CCF. The fistula was closed successfully. After the procedure, the case developed 6th cranial nerve palsy and on the 27th follow-up month; although on a decreasing trend, the symptoms still persist.

Case 2 (Case number 22 L CCF): The case underwent coil embolization via IPS for type C CCF. The fistula had closed in the first session, and the residue supplied by the ECA deep temporal artery ends persisted on the follow-up DSA at the second month. In the second session, Onyx was performed via the ECA. After the second treatment session, the case developed a thalamic microinfarction.

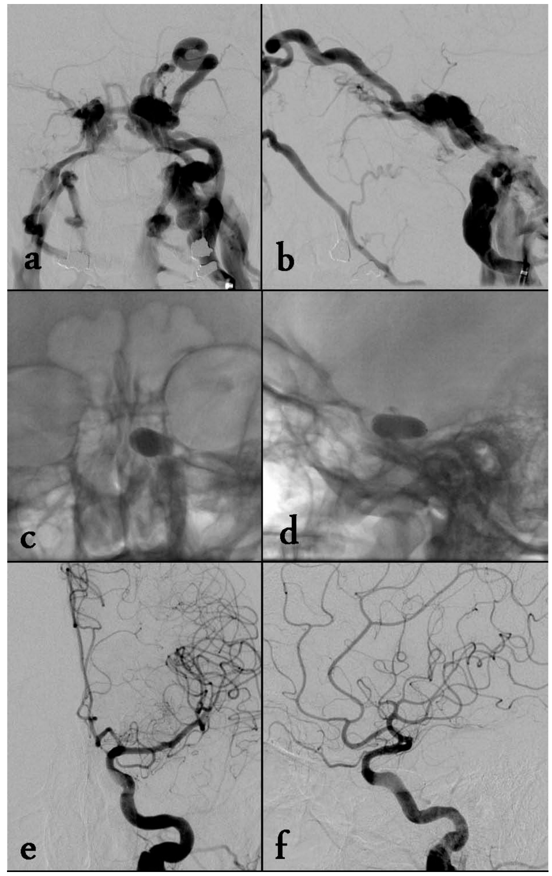

Figure 2. Barlow type a CCF (case no. 19) the rupture of the ICA to cavernous sinus, was seen at the left ICA selective angiography (a) and (b) of the 45 years old female patient who admitted after head trauma with the symptoms of chemosis, severe orbital pain-pressure sensation in her left eye, and flow in the distal part was interrupted. Venous drainage has occurred to the bilateral superior opthalmic veins and inferior-SPSs. in the treatment, detachable balloon was implemented transarterially via ICA (c) and (d). Postperative control angiography showed recanalization of the distal circulation and total closure of the fistula tract (e) and (f).

All of the patients underwent clinical follow-ups. The mean follow-up range was 2–77 months (23 ± 53). Complete clinical recovery was achieved in 20 of the 23 patients. Despite a decrease in the symptoms that were present before the procedure, diplopia persisted in one case, chemosis in one case, and ptosis persisted in one case.

Discussion

In accordance with other series, our study shows that endovascular CCF treatment has high success and low complication rates [1,3,6,7]. Most cases were treated

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successfully in one session, and in a small number of cases, treatment was achieved successfully in a maximum of two sessions. Relatively long-term clinical and angiographic follow-up results demonstrate the success of endovascular treatment.

Direct and indirect CCFs develop with different mechanisms, and thus, the symptoms develop with different velocity and severity. Rupture of ICA opening into the cavernous sinus after severe head trauma and saccular ICA aneurysm rupturing into the cavernous sinus are the two main mechanisms behind direct CCFs. Therefore, the symptoms are acute and severe [7,8]. Four of our direct CCF cases developed secondary to the trauma and one case developed secondary to the aneurysm rupture. The mechanism behind indirect CCFs is the growth of dural branches in ICA and ECA arterioles on the cavernous sinus wall, during the cavernous sinus recanalization after a potential cavernous sinus thrombosis [7].

When the fistula track and cavernous sinus are small, coil embolization is mostly adequate and successful [9]. Among five of our cases with direct CCFs, three cases with small fistula tracks were treated with coils (case number 11) and stent + coil combinations (cases number 7 and 14). The main purposes of stents usage were uncertainty in dissection and preserving the parent artery patency during endoluminal reconstruction with coiling [6]. The stents we used in the two cases targeted to prevent the coil migration.

If the ICA morphology was suitable to advance the balloon, detachable balloons could be used, especially in cases with wide fistula tracks and cavernous sinuses [8]. These balloons have been used widely and successfully in the treatment of CCFs for many years [10,11]. The success of balloon embolization has been reported as 88%–99% [12,13]. The only case was obliterated in one session with a detachable balloon (case number 19) was suitable for balloon access with fistula morphology. In the long-term DSA follow-up, the fistula was closed and the case was asymptomatic.

Covered stents were quite successful in severe traumatic CCFs with arterial lacerations. However, coated stents had disadvantages such as being hard, having low traceability, and wide diameters. In addition, the long-term results of these stent grafts were not known enough [6]. In one of our traumatic CCF case (case number 10), a covered stent was selected due to the dramatic tear in the ICA. According to the results of the postprocedural DSA and the three-month follow-up, complete recovery was achieved in this patient.

In most series, direct CCFs were treated via the arterial route [7,14]. In the series of Barry *et al*. [7], the arterial route was used in 9 (ICA), the venous route was used in 2 (IPS), and a combined approach was used in 1 of the 12 direct CCF cases. Xiaojian *et al*. [14] successfully treated 32 direct CCF via arterial route with different embolization techniques. The arterial route (ICA) was used in four and the venous route was used in one of our five direct CCF cases.

Indirect CCFs usually cause severe clinical symptoms and rarely close spontaneously. Hence, they have to be treated. Detachable coils are used successfully in the treatment of indirect CCFs [6]. The ability to retrieve, the chemically inert platinum structure, and the lack of neurotoxic effects on the nerves inside the cavernous sinus are the main advantages of coils. Therefore, we used detachable coils in most of our cases. The main disadvantages of detachable coils are their high cost. Placing coils also requires repeated DSA implementation and hard to evaluate the parent artery patency, because they are highly radiodense [6,8]. However, the use of biplane angiography has partially reduced these disadvantages. In addition, using a large number of coils to achieve successful embolization may cause compression in the cavernous sinus and cranial nerves [6]. In one of our cases (case number 15), sixth nerve palsy developed after coil embolization. In particular, if the fistula track is wider than the cavernous sinus, the procedure can be accompanied by stent modeling to prevent the coils from leaning into the parent artery [6,8]. We did not require stent modeling in any of our indirect CCF cases.

Liquid embolization agents can be used alone or in combination with coils. In the Barber *et al.* [1] series, during the mean 12.4-month angiographic follow-up after treatment with liquid embolization agents, no relapses were observed which require any recanalization or advanced treatment. Among these, n-BCA is preferred for its low cost and due to the fact that it forms thrombosis rapidly in the fistula tract; however, if it leaks into the ICA, it may cause catastrophic complications [8]. Onyx is a more expensive embolization agent; however, nonadhesive characteristic makes it important that allows it to be injected in a small amount over a longer period without sticking to the catheter and allows complex fistulas to be treated in one or a few sessions [6]. We successfully treated one (case number 9) of our type D indirect CCF patients via the venous route with n-BCA, and the patient was asymptomatic and stable at the three-month follow-up.

It has been reported that the use of Onyx and n-BCA materials with coils contributes to complete obliteration with an attachment closure at the center of the coil mass [6]. One of our cases (case number 22 L CCF) with indirect CCF was treated with a coil via the venous route in the first session. However, since a full clinical recovery was not accomplished and a residue supplied by the ECA was seen on the angiographic follow-up, the end branch of this artery was entered superselectively and the residual fistula was treated successfully.

The largest risk of using liquid agents is the possibility of retrograde penetration of the embolization agent to the feeding artery, which may cause catastrophic complications distally [8]. In the case, we treated with Onyx, a millimetric thalamic infarction developed after the procedure and it was confirmed with an MRI; however, the patient was stable neurologically and discharged without any problems.

The majority of our cases with indirect CCFs were treated via the venous route toward other series in the literature [1,2]. One of these cases (case number 23) had a rather wide feeding artery with an adequate angle (MHT) and the fistula was closed through an arterial approach with the same route. The other case (case number 22 L CCF) was only partially closed at first session via the venous route and this was the case we treated by the arterial route (ECA end branches) with Onyx.

In the two cases with type D and type B indirect CCFs, we were unable to access the cavernous sinus via the venous route, probably due to the IPS and SPS veins being closed. We were also unable to succeed in arterial approach. These cases were treated successfully in cooperation with ophthalmic surgery by catheterizing the SOV with direct orbital puncture. The patients were asymptomatic in long-term clinical follow-up at the 24th and the 52nd months, respectively. SOV entrance carries risks such as optic nerve or globe damage, intraorbital hematoma, hemostasis difficulties due to arterialized flow, and it should be reserved for use in conditions when the cavernous sinus cannot be accessed via other routes [8]. No orbital complications developed in either of our two cases.

Complications related to the procedure developed in only two of our cases. In one of type D CCF case (case number 15) treated with coil embolization, sixth cranial nerve palsy developed after the procedure and persisted. It probably developed secondary to the use of many coils to achieve successful embolization. The other two cases were embolized with coils via the venous route but underwent retreatment with Onyx via the arterial route in a second session due to the residue. Later, a thalamic microinfarction developed; however, the case recovered neurological near completely. Barry *et al.* [7] reported two thromboembolic stroke complications in their series with a similar number of patients.

The main limitations of the study were the retrospective method, the limitation of the cases, and the content of heterogeneous cases.

Finally, the current approach in CCF treatment is endovascular treatment using coils, liquid embolization agents, detachable balloons, stents, and combinations of these methods. Parallel with current technology, the endovascular techniques and materials used are changing and developing rapidly. The critical point in the treatment is achieving complete fistula obliteration in the least possible number of sessions with appropriate embolization agents via the appropriate route. Endovascular treatment which performed with use of appropriate technique and material has high success and low complication rates. Although CCF and its treatment is a known antity, we believe that our cases which had long-term results will contribute to the literature.

Acknowledgements

None.

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