LITERATURE REVIEW

Endovascular Coiling Embolization in Direct Carotid-Cavernous Fistula

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ABSTRACT

Background: Carotid cavernous fistula (CCF) is a rare life-threatening. Rapid endovascular technical development has provided as a safe and effective treatment strategy for direct CCF (DCCF). Coiling treatment is a promising endovascular procedures solution in DCCF.
Method: A comprehensive PubMed, Clinical key, and Ophthalmology database search was conducted using appropriate keywords (direct carotid cavernous fistula, endovascular treatment, embolization, and coil). Inclusion criteria was only article in English.
Results: Nine studies (8 case series and 1 cohort study) were found suitable. From these results, endovascular coiling treatment can close the fistula with radiological evaluation and improvement in clinical symptoms in most of studies (90% and 88%, respectively). Post-operative and long-term complication were reported in some studies.
Conclusion: Efficacy of endovascular coiling embolization appears promising in management of DCCF. There were no intra-operative complications and post-operative complications, such as unraveled micro-coil. The recurrence fistula rate of 4% is common in fistulas with larger tears associated with a larger CCF.

Keywords: *direct carotid cavernous fistula, endovascular, treatment, embolization, coil*

Carotid cavernous fistula (CCF) is a rare life-threatening. Direct carotid-cavernous fistulas (DCCF) are an anatomical classification of CCF that showed by abnormal arteriovenous shunts between the internal carotid artery (ICA) and the cavernous sinus. They progress rapidly, necessitating urgent treatment. Traumatic is the most common etiology of DCCF, which accounting for 80%.[1-5] Direct carotid-cavernous fistulas (DCCF) often present acutely with classic triad (includes pulsatile exophthalmos, orbital bruit, and conjunctiva injection). Majority patients suffer orbital disturbances, such as ophthalmoplegia, diplopia, proptosis, high IOP, orbital pain due to cavernous sinus hypertension and eye muscles edema caused by venous congestion. Venous hypertension on intracranial cortical veins can lead to subarachnoid hemorrhage which occurs in 5% patients and 1-2% manifest life-threatening epistaxis.[4] Cerebral angiography is the gold-standard imaging modality in the diagnosis of CCFs.[3]

The goal of CCF treatment is to completely occlude the fistula while preserving the normal flow of blood through the ICA. Conservative, surgical or endovascular therapy can be performed to closure management.[6] Endovascular therapy using coiling has grown wider and prefer used by interventionist compared with ballooning, liquid embolic agents or covered stent graft as their reliable and controlled deployment. Coiling treatment

are promising endovascular procedures in DCCF.[2,6,7,8]

The exact method chosen in each case depends on the anatomy of the fistula and operator/institutional preferences. Many interventionists prefer to use detachable platinum coils because of their reliable and controlled deployment. The coils can be adjusted easily or even removed if the placement is not optimal. Regarding of these options, ophthalmologist would like to know the efficacy and disadvantage of coiling technique of endovascular procedure based on evidence literatures.

METHODS

A comprehensive search via PubMed, Clinical Key, and Ophthalmology Advance using search terms "direct carotid cavernous fistula", "endovascular treatment", "embolization", and "coil". No limitation was set regarding number of subjects, year of publication and type of study. Only articles in English were included. Reviewing abstracts and choosing relevant study were performed first. From these selected articles, full-text analysis was carried out from these selected articles. The articles show about efficacy and disadvantage of the coiling technique of endovascular procedure based on evidence literature, include fistula closure, clinical improvement symptoms, complication, long-term outcome, and duration of new clinical signs after the system in the long term outcome.

RESULTS

The process of database search is presented in Fig 1. All selected studies were classified based on level of evidence of Oxford Centre for Evidence-based Medicine.[9] In abstract result:Nine studies (8 case series and 1 cohort study) were found suitable.

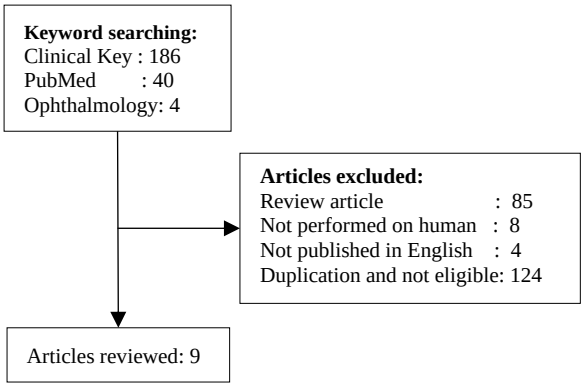

Fig 1. Flowchart of study selection process

Studies that were reviewed in this article were published from year 2001 to 2017 with majority of case series (level 4 of evidence). We only evaluate coiling technique as endovascular treatment on DCCF in all studies. Total patient included in this review is 117 patients. All articles are available in full text. Articles summary are presented in table 1.

Wide range of age was identified (25.5-53.7 years). The shortest mean follow-up duration was about 6 months, while the longest was 4.5 years. All studies revealed male as dominating in gender, with overall ratio ± 2:1 compared to female. Most cases are traumatic DCCF, only 7 patients arise spontaneously. Some studies showed that duration of first symptoms until elective procedure were 5,96 - 12.5 months. Clinical symptoms that arise in nine studies were shown in figure 2. Result of endovascular coiling embolization and complications are presented in table 2 and table 3, respectively.

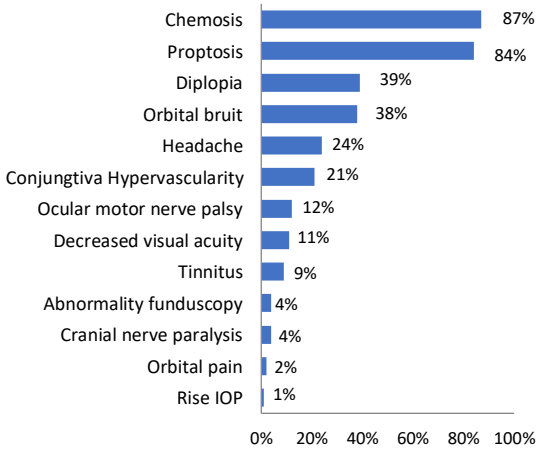

Fig 2. Clinical Symptoms of DCCF

Table 1. Articles Summary

No	Authors	Place of Study	Year	Level of evidence	Sample Size	Follow Up (mean)
1.	Ghannam AB, et al[10]	Colorado	2017	4	2	4,5 yr (4-5 yr)
2.	Joshi Kc, et al[12]	India	2016	4	15	1 yr
3.	Hassan T, et al[13]	Egypt	2015	4	19	6 mo and 5 yr
4.	Stephan S, et al[14]	France	2015	4	10	10 ± 14 mo (6-36 mo)
5.	Zhang Z, et al[8]	China	2013	4	15	22 mo (14-48 mo)
6.	Renzis AD, et al[15]	Turkey	2013	4	13	3.8 yr (6 mo-6 yr)
7.	Mahmoud M, et al[16]	Egypt	2013	3	12	6 mo
8.	Luo CB, et al[11]	Taipei	2013	4	24	19 mo (3-48 mo)
9.	Luo CB, et al[17]	Taiwan	2006	4	7	16 mo (8-25 mo)

Abbrevations; *yr: years; mo: month*

Table 2. Result after endovascular coiling embolization in radiology evaluation & symptom

No	Authors	n	Result								Follow Up
			Radiology			Signs & Symptoms				Lost to follow up	
			Complete fistula occlusion	Partial fistula occulusion	Unable occluded	Symptom Resolved	Improved	Worsen	Persistent symptoms		
1.	Ghannam AB, et al[10]	2	2	-	-	2	-	-	-	-	4,5 yr (4-5 yr)
2.	Joshi Kc, et al[12]	15	12	1	2	12[a]	-	-	3[a]	-	-
3.	Hassan T, et al[13]	19	16	3	-	18	-	1 (covered stent)	-	-	6 mo and 5 yr
4.	Stephan S, et al[14]	10	4	-	-	1	2	-	1 (optic neuropathy sequele)	6	10 +/- 14 mo (6-36 mo)
5.	Zhang Z, et al[8]	15	14	-	1 (balloon)	14	-	-	-	-	22 mo (14-48 mo)
6.	Renzis AD, et al[15]	13	9	-	-	8	-	-	1 (partial n. III palsy, bilateral pupillary light reflex loss and moderate ophthalmoplegia)	4	3.8 yr (6 mo-6 yr)
7.	Mahmoud M, et al[16]	12	12	-	-	12[b]	-	-	3 (diplopia & n. VI palsy)	-	6 mo
8.	Luo CB, et al[11]	24	21 (n=4 recurrent)	4 (n=4 retreatment)	-	21	-	-	-	-	19 mo (3-48 mo)
9.	Luo CB, et al[17]	7	7	-	-	7	-	-	-	-	16 mo (8-25 mo)
TOTAL		**107**	**97/107**	**8/107**	**3/107**	**95/107**	**2/107**	**1/107**	**9/107**	**10**	
%			**(90%)**	**(7%)**	**(3%)**	**(88%)**	**(2%)**	**(1%)**	**(8,4%)**		

Abbrevations; *(-): not mentioned; a: disappearance of orbital bruit*; *b: Headache, proptosis and chemosis disappeared*

Table 3. Complication after endovascular coiling embolization

No	Authors	Complication				Duration of new symptoms after procedure	Average number of micro-coils
		Intra Operative	Post-Operative	Recurrence	Long-term Complication		
1.	Ghannam AB, et al[10]	-	-	-	6th nerve palsy and 3rd nerve palsy. Delayed onset diplopia after succesfull treatment	4/5 yr	16 coils
2.	Joshi Kc, et al[12]	-	-	-	-	-	-
3.	Hassan T, et al[13]	-	-	-	Pseudoaneurysm	3 yr follow up	
4.	Stephan S, et al[14]	-	-	-	Optic neuropathy sequele	-	-

5.	Zhang Z, et al[8]	-	Complete occlusion with loose packing. Microcoil unraveled	-	-	-	5 (2-16 coils) L: 0.95 (0.12-1.86 m)
6.	Renzis AD, et al[15]	-	-	-	Pseudoaneurysm	-	-
7.	Mahmoud M, et al[16]	-	-	-	-	-	-
8.	Luo CB, et al[11]	-	-	4 patients	6th nerve palsy and 3rd nerve palsy	3 mo (recurrent and residual)	14 (2-31 coils) L:1.89 (0.16-7.56 m)
9.	Luo CB, et al[17]	-	-	-	Pseudoaneurysm	-	7.7 (2-13 coil) L: 0.9 (0.16-2.34 m)

***Abbrevations; (-)**: not mentioned; yr: years; mo: months L: length*

DISCUSSION

The most commonly used classification scheme established by Barrow divides the CCF into four types, depending on the arterial supply.[1] Type A or direct CCF is the most common type accounting for up to 80% of all CCF and mostly because of a tear in the carotid wall after trauma.[3] In this review study, showed that DCCFs are dominant in male that female and mostly caused by trauma. Spontaneous DCCFs are more common in older women and usually caused by the rupture of a cavernous aneurysm or by the spontaneous rupture of a congenitally weakened, atherosclerotic, or diseased artery.[1,3] The clinical feature of CCF is a direct consequence of elevation in intra-cavernous pressure. This pressure is transmitted anteriorly to the ipsilateral orbit and posteriorly to the inferior petrosal sinus. Anterior drainage leads to orbital vein congestion and transudation of fluids, increased intraocular pressure, impaired retinal perfusion, and rupture of dilated veins. The most frequent complaints are in the orbital region as the classic triad of exophthalmos, conjunctiva chemosis, and cephalic bruit.[1,3] In this review, clinical signs and symptoms include chemosis (87%), proptosis (84%), diplopia (39%), orbital bruit (38%).

Management with endovascular coiling treatment can close the fistula through radiological evaluation in 90% of the time. However, 3% cases are unable occluded and require further management. Joshi KC et al reported had 3 patients with incomplete occlusion of fistula, which is possibly due to multiple fistulous entry points or severe laceration of the carotid artery, which are not accessible to the micro catheter. The availability of suitable intracranial stents may be performed to treat these difficulty.[15] Zhang Z et al reported that one patient with fistula and parent artery were occluded with balloons because there were a large residual flow after successful 5 coils deployment.[8]

Improvement in clinical symptoms rate is 88%. The persistent complaints were optic neuropathies, cranial N.III, and N.VI palsy. Stephan et al reported all patient had vision recovery except one patient persistently sustain optic neuropathy with very low vision.[14] Luo CB et al reported recurrence rate were 4% which is particularly common in larger tears of fistulas associated with larger CS as less coil packing density compared with small fistula and small CS.[17] After complete closure of a CCF, Joshi KC et al reported that chemosis and proptosis generally resolve in hours to days. Cranial nerves palsy typically resolve after six weeks. The degree of vision recovery prior to intervention, largely depend on the pathogenesis, severity, and duration of the pre-intervention deficit.[15]

The complications found in several studies were post-operative, i.e. a complete occlusion with loose packing and the unraveled micro-coil. In order to prevent the retrograde herniation of the embolic material into parent artery and distal intracranial circulation, the assistance of a

non-detachable balloon (balloon-assist technique) or a porous stent may be chosen especially in the setting of a large tear in the ICA. The long term outcomes after successful endovascular coiling embolization are the presence of pseudoaneurysm, N.VI palsy and N.III palsy, and optic neuropathy. Ghannam AB et al reported a delayed onset of diplopia after successful treatment is still unknown. Late compression of cranial nerves in cavernous sinus due to coil mass may cause chronic ischemia. Delayed inflammation due to thrombophilic nidus created by coil mass or injury to the cranial nerves manifests later due to decompensated strabismus. Other studies also explored the use of the number of micro-coils used in the study. The number and length of coils largely depended on the size of the CS.

The limitations from this studies not only the sample size and level of evidence due to the nature of this DCCF, but also most of the studies didn't mention the duration of symptoms until the elective procedures and the duration of new symptoms after endovascular coiling embolization.

CONCLUSION

Efficacy management of DCCF with endovascular coiling treatment appears promising to improve the clinical symptoms and reached complete fistula occlusion. There were post-operative complications, such as unraveled micro-coil. The long-term outcomes are pseudoaneurysm, 6th nerve palsy, 3rd nerve palsy, and optic neuropathy. The recurrence fistula rate of 4% is common in fistulas with larger tears associated with a larger CS. These long-term outcomes were happened in the range from 3 months to 5 years after the procedure. Future studies in larger sample size are still required.

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