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Case Report

Treatment of Carotid Cavernous Fistula Presenting with Contralateral Exophthalmos: Several Experiences of Graft Stent

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Objective: Graft stent for carotid cavernous fistula (CCF) is known to be a useful treatment. Here, we report our experiences of treatment with graft stents for CCFs.

Case Presentations: From 2015 through 2018, six patients underwent graft stent placement for CCF occlusion. Clinical and angiographic data were retrospectively reviewed. Access and deployment of a graft stent was successful in all patients and complete occlusion immediately after the procedure or 3-month follow-up angiography.

Conclusion: Graft stents should be considered as an alternative option of treating CCFs and preserving the parent artery by arterial wall reconstruction especially in patients with a fistula that cannot be successfully occluded with detachable balloons or coils.

Keywords ▶ carotid cavernous fistula, graft stent, contralateral exophthalmos

Introduction

Endovascular occlusion using detachable coils or balloons is one of the established treatments for carotid cavernous fistulas (CCFs).[1,2] In recent years, endovascular treatment using graft stents provides good results in terms of less invasive occlusion of fistulas with preservation of the parent artery.[3–6] In the present study, we reported a case with contralateral symptom and five cases with ipsilateral symptoms treated by graft stents.

Patient population

From January 2015 through December 2018, a total of six cases of CCF were treated by the endovascular covered stent grafting at our institution. Two were direct CCFs with a symptom triad and four were indirect CCFs with diplopia or exophthalmos. For indirect CCF case, stent graft is considered when three conditions are satisfied: (1) no effect on manual compression, (2) it is impossible to treat Gamma knife, and (3) the origin is not clear, but the fistula site is observed in the cavernous portion. These patients had periodic clinical follow-up examinations (at 0–33 months) with all receiving angiographic follow-up examination (at 0–15 months). A Graftmaster (Abbott Vascular, Santa Clara, CA, USA) stent graft was applied in all patients with CCF. A graft stent was applied in five patients primarily as the first choice and other one as an alternative option after incomplete occlusion with stent-assisted coil embolization. Clinical and angiographic features are summarized in **Table 1**.

All patients were loaded with plavix 300 mg prior to the procedure, and endovascular procedures were performed under general anesthesia. The angio-system was evaluated using the Axiom Artis dBA Bi-plane angiographic system™ (Siemens medical system, Munich, Germany). After placing 6 Fr. guiding catheter in proximal internal carotid artery (ICA), a microcatheter was passed fistula segment of ICA, and a Graftmaster coronary stent graft was hand mounted onto an angioplasty balloon. Intravenous heparin was used during all cases and low molecular weight heparin was used for 5 days and dual antiplatelet medication was maintained for at least 6 months after the procedure.

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Table 1 Data of patients with CCF treated with graft stents with follow-result

Patient sex/ age (years)	Signs/ symptoms	CCF type	Stents sizes	Post-procedural angiographic result	Final angiographic F/U
F/73	Proptosis, visual disturbance	Indirect	Graftmaster 3.5 × 16 mm	Radioanatomic cure without complications	Stable CCF occlusion with ICA patency
*M/28	Conjunctival injection, mild proptosis	Direct	Graftmaster 4.5 × 16 mm	Small endoleak was observed	Stable CCF occlusion with ICA patency
†M/65	Proptosis, visual loss	Indirect	Graftmaster 4.0 × 12 mm	Radioanatomic cure without complications	Stable CCF occlusion with ICA patency
M/58	Visual disturbance, bruit	Indirect	Graftmaster 4.8 × 16 mm	Radioanatomic cure without complications	Stable CCF occlusion with ICA patency
F/70	Headache, bruit	Direct	Graftmaster 4.8 × 16 mm	Small endoleak was observed	Stable CCF occlusion with ICA patency
F/64	Contralateral exophthalmos, visual disturbance, bruit	Indirect	Graftmaster 4.5 × 19 mm	Small endoleak was observed	Stable CCF occlusion with ICA patency

*Complete obliteration at immediately angiography, but small endoleak was observed follow-up angiography after 3 months. †Graft stent insertion was performed after stent-assisted coil embolization. CCF: carotid cavernous fistula; ICA: internal carotid artery

As a general rule, 6 months after the procedure, follow-up tranfemoral femoral cerebral angiography (TFCA) was performed as in the case of usual stent-assisted coil cases, and in some case of endoleak, TFCA was performed after 3 months.

Placement of graft stent was technically successful in all six patients. After stent placement, immediate complete occlusion of the fistula was obtained in three cases and near complete occlusion with small endoleak was observed in three patients. The patency of the ICA was preserved in all patients. CCFs-related symptoms disappeared within 1–14 days after treatment in all patients without thromboembolic events. There was no mortality or morbidity related to the procedure. Follow-up angiography showed complete occlusion of CCFs in three patients including the two patients with small endoleak on postoperative angiography. However, recurrence was seen during follow-up in one patient in whom immediate complete exclusion on postoperative angiography. This patient received re-dilation of the stent using a balloon. Final follow-up angiography showed complete disappearance of all CCFs and patency of the stents without in-stent stenosis.

Case Presentations

Case 1

A 64-year-old woman was admitted to an outside institution for presenting with left-sided decreased vision, exophthalmos, injection, and periorbital bruit. She did not have a history of head trauma, and symptoms gradual got worse 2 months ago. Brain CT and TFCA (**Fig. 1**) were performed, as a result, Rt. CCF was observed. As a result of angiography, the fistula was located at the genu portion of the cavernous ICA. Diameter of fistular portion of ICA was 4.6 mm and the origin of fistular point was not clearly observed. Graft stent was placed to the fistula location and repeated balloon dilatation was performed with gradually increasing pressure. As a result of immediately post-procedural angiography, small endoleak was found. After confirmation of ICA patency and absence of thrombus, the procedure was completed. After 3 months, endoleak was not observed on F/U angiography and CCF was complete obliteration (**Fig. 2**).

Case 2

A 28-year-old man was visited to our emergency department for presenting with visual disturbance, mild proptosis, and Lt. conjunctival injection. He had a surgical history at 8 months ago with cerebral contusion with skull base fracture after traffic accident. Brain computed tomography angiography (CTA) (**Fig. 3**) was performed, as a result, Lt. CCF was observed. We were planned for endovascular treatment and performed angiography. As a result angiography, it revealed profound left CCF draining to ophthalmic vein, inferior petrosal sinus. The diameter of fistular potion of ICA was 4.8 mm. Endovascular graft stent apply was performed (**Fig. 3**). 6 Fr. guiding catheter was placed in left proximal ICA and 14 microwire was passed at left cavernous ICA segment and graft stent was applied at fistular location and repeated balloon dilatation was performed with 18 atm pressure. As a result of immediately post-procedural angiography, nearly complete resolve of CCF, but small endoleak was found (**Fig. 4**). After 6-month follow-up angiography, there was a complete obliteration of CCF and endoleak was not observed anymore (**Fig. 5**).

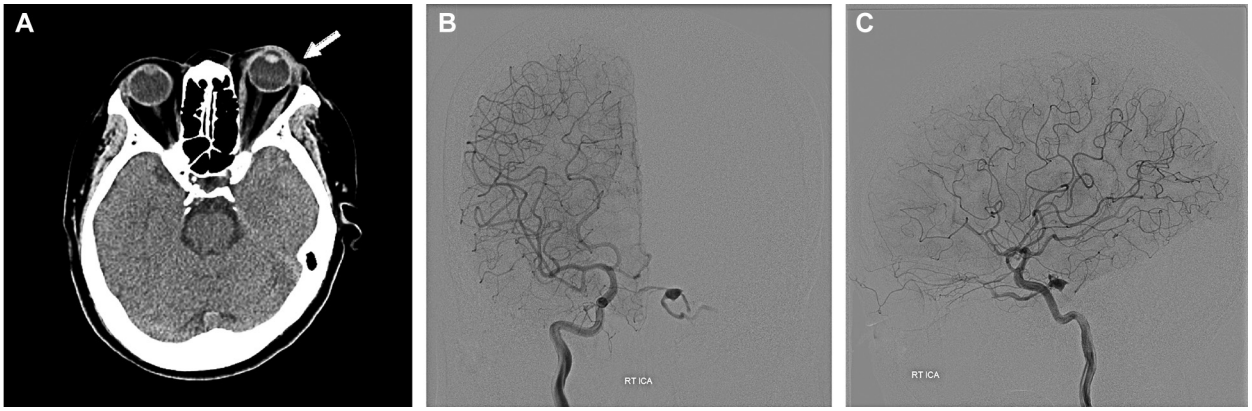

Fig. 1 On Brain CT (**A**) and diagnostic angiography (**B** and **C**), Lt. exophthalmos (white arrow) was identified at brain CT and Rt. cavernous ICA-Lt. cavernous sinus fistula was found at angiography. CT: computed tomography

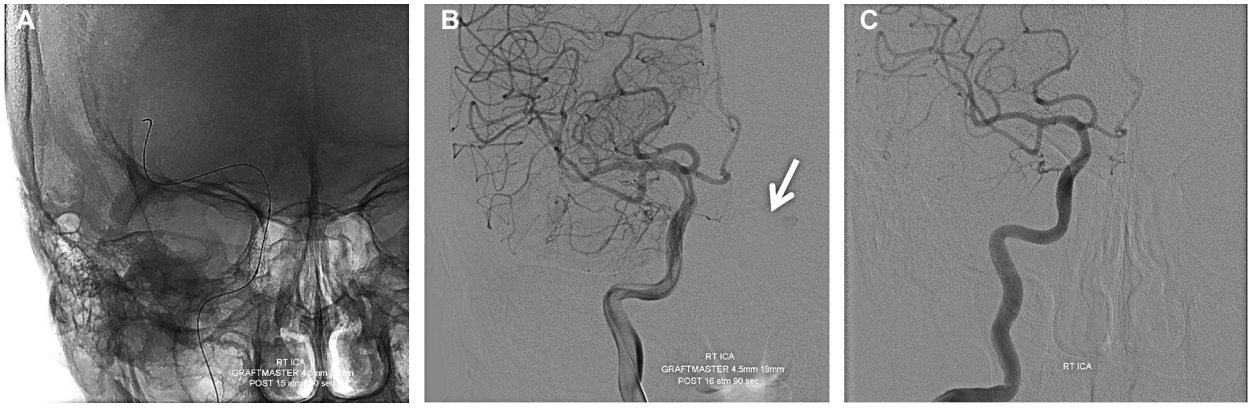

Fig 2 (**A**) Graftmaster™ covered stent apply, (**B**) immediately post-procedural angiography. CCF was nearly complete obliteration but small endoleak was found (white arrow), and (**C**) 3-month F/U angiography. Endoleak was not found and complete obliteration of CCF. CCF: carotid cavernous fistula

Fig. 3 CT angio (**A** and **B**) and diagnostic angiography (**C** and **D**) image. Lt. CCF was found and draining to Lt. IPS and dilatation of Lt. ophthalmic vein. It also shows delayed intracerebral blood flow. CCF: carotid cavernous fistula; IPS: inferior petrosal sinus

Discussion

Recently, treatments of CCFs using coils or balloons are common, but these treatments may result in parent artery occlusion or cranial nerve palsy due to over packing. There is another potential risk of coil dislodgment into the ICA, especially if the defect is large.[7] In addition, detachable coils be an extremely expensive option.[8] On the other hand, detachable balloons are unreliable due to unexpected deflation resulting in recurrence or bleeding.[2]

Recently, graft stents have been used for the treatment of cerebral aneurysms, arterial dissections, and arteriovenous fistulas as well. Several reports have been published about the feasibility, safety, and clinical outcomes of graft stents in the treatment of CCF, and presenting that a graft stent can be an effective method for treatment of CCF.[9–12]

Fig. 4 Procedural (**A**) and immediately post-procedural (**B** and **C**) angiography. Near total obliteration of Lt. CCF but small endoleak was found. CCF: carotid cavernous fistula

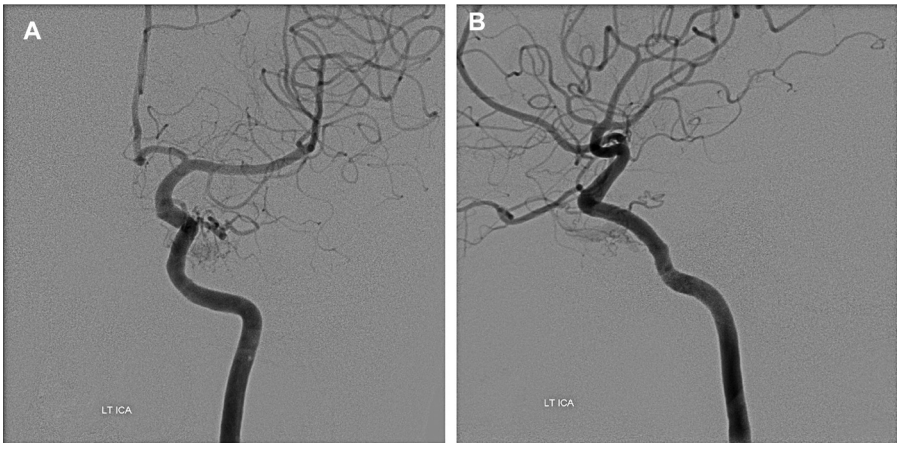

Fig. 5 After 6-month follow-up angiography (**A** and **B**), endoleak was not found and complete occlusion of CCF. CCF: carotid cavernous fistula

In 2001, Weber et al.[13] and Kocer et al.[14] reported each one case of direct CCF successful treated with graft stent. Since then, research on the effectiveness of graft stents on direct CCF has been published, but the research of indirect CCFs treated by graft stent is difficult to find. In our cases, we performed a treatment of indirect CCF in four cases and had confirmed to successful outcome.

Endoleak is defined as a persistent perfusion of the space between the stent graft and the parent vessel wall. This is the most common cause of procedural failure in this technique.[12] Hoit et al.[15] reported their experience with a covered stent for direct carotid cavernous fistula (DCCF) and a traumatic pseudoaneurysm. According to the report, transient endoleak occurred in 83% (5/6); the endoleaks were related to poor stent vessel apposition or size mismatch. In our cases, transient endoleaks were found in 50% (3/6) immediately after stent deployment. However, in all cases, endoleak disappeared and CCF was completely obliterated on follow-up angiography after 3–6 months of the procedure.

Graft stent apply for CCF has several advantages over coil embolization or stent-assisted coil embolization. Graft stent apply is advantageous for maintaining patency of ICA, also unlikely to recanalize CCF due to coil compaction. In addition, effective results can be expected even with very small fistula locations. However, there are some caveats for graft stent apply for CCF. First, the ipsilateral intracranial circulation is preserved in patients with good collateral flow even after ICA occlusion. Second, when fistulas are located in a less a tortuous portion of the carotid artery, accurate positioning of the stent is possible. Third, if no critical side branches adjacent to the site of the fistula, safe placement of the graft stent is possible.[16]

Our results suggested that this treatment strategy was feasible and efficient and that it should be considered as an

alternative treatment especially when the use of detachable balloons or coils were difficult to occlude the fistula with preserving the ICA patency.

Conclusion

When a CCF cannot be safely occluded with detachable coils or balloons, graft stents would be an option to occlude CCFs with preserving the parent artery. Although a larger sample and expanded follow-up are needed, our series shows that covered stents can be used in the treatment of CCFs with symptomatic relief as experience with this device increases, materials continue to improve, and more data are accumulated.

Disclosure Statement

All remaining authors have declared no conflicts of interest.

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