

Abstract

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SURGICAL NEUROLOGY

Surgical Neurology 72 (2009) 169-174

www.surgicalneurology-online.com

Arteriovenous Malformation

Combined covered stent and onyx treatment for complex dural arteriovenous fistula involving the clivus and cavernous sinus Zhong-Song Shi, MD, PhD^{a,b}, Tie-Wei Qi, MD^a, Nestor R. Gonzalez, MD^{b,c}, Jordan Ziegler, MD^b, Zheng-Song Huang, MD, PhD^{a,*}

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Received 2 August 2007; accepted 5 March 2008

Background: Complex DAVFs involving both the clivus and cavernous sinus are rare, especially when associated with brainstem compression from a large varix. In this report, we describe the use of a covered stent in combination with a liquid embolic agent to cure a complex clival-cavernous DAVF.

Methods: A 46-year-old man presented with 6 months of dizziness, dysphagia, and progressive dysarthria. Magnetic resonance imaging showed tortuous and enlarged right cavernous and preclival flow voids. There were also bilateral preportine varices compressing the ventral pons, which led to marked dorsal pontine edema. A cerebral angiogram revealed a clival DAVF supplied by multiple branches of the right ECA, as well as the MHT of the right ICA.

Results: An endovascular cure was achieved by deploying a covered stent in the right cavernous ICA, followed by transarterial embolization of the feeding arteries originating from the ECA with Onyx (ev3, Irvine, Calif). This combined approach resulted in complete occlusion of the fistula. His 1-month follow-up angiogram confirmed persistent occlusion of the fistula and preserved patency of the right ICA. The patient made a full recovery without any new symptoms, and he remained neurologically intact at 18-month follow-up.

Conclusion: The combined technique of covered stent placement and Onyx transarterial embolization is valuable for the management of complex DAVFs supplied by branches of both the external and internal carotid arteries.

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Keywords: Dural arteriovenous fistula; Endovascular therapy; Covered stent; Liquid embolic material; Onyx

1. Introduction

Despite ongoing progress in surgical and endovascular techniques for the management of cerebrovascular diseases,

the therapeutic options for certain types of DAVFs remain challenging [2,3]. Dural arteriovenous fistulae involving both the clivus and cavernous sinus are rare and complex [16], especially when associated with large varices, which can cause significant mass effect and brainstem edema. Although there have been technical case reports of covered stent placement in the cavernous carotid artery for aneurysm exclusion [4,6,12,14], we now describe a technical case report of combined covered stenting and transarterial embolization with a liquid embolic agent for cure of a complex dural fistula involving both the clivus and cavernous sinus.

Abbreviations: DAVF, dural arteriovenous fistula; ECA, external carotid artery; ICA, internal carotid artery; MHT, meningohypophyseal trunk; MR, magnetic resonance; MRI, magnetic resonance imaging.

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^{0090-3019/\$ –} see front matter @ 2009 Elsevier Inc. All rights reserved. doi:10.1016/j.surneu.2008.03.022

2. Case report

2.1. Clinical presentation and angiographic features

A 46-year-old man presented with a 6-month history of dizziness, dysphagia, and progressive dysarthria. His neurological examination revealed partial palsies of cranial nerves III, IV, and VI. Magnetic resonance imaging of the brain revealed prominent and tortuous signal voids in the right cavernous sinus, bilateral prepontine cisterns, cerebellopontine cistern, lateral cerebellomedullary cistern, and the right pontocerebellar angle region. Two giant prepontine varices, measuring 25×18 mm on the right and 23×20 mm on the left, were noted to compress the ventral surface of the pontomesencephalic region, causing marked brainstem edema (Fig. 1A). Cerebral angiograms demonstrated a high-flow DAVF involving the clivus and right cavernous sinus. There was complex arterial inflow, and the fistula was fed by multiple branches of the right ECA, including the middle meningeal artery, accessory meningeal artery, artery of the foramen rotundum, and the occipital artery. Multiple tiny branches of the MHT of the right ICA also supplied the DAVF. The venous drainage was primarily into the right cavernous sinus with retrograde flow into the right lateral pontine vein, transverse pontine vein, left lateral pontine vein, left superior petrosal sinus, and the left transversesigmoid sinus. There was also relatively minor deep venous drainage via the lateral mesencephalic vein, basilar vein, and the vein of Galen into the straight sinus (Fig. 2).

2.2. Endovascular procedure

During the procedure, heparin was administered systemically to keep the activated clotting time between 250 and 300 seconds, and Tirofiban hydrochloride, an antiplatelet glycoprotein IIb/IIIa antagonist, was infused intravenously during the procedure. We performed a preprocedural balloon test occlusion at the petrous right ICA for 30 minutes, anticipating the possibility of failed covered stent deployment, with potential necessity for ICA sacrifice. The patient tolerated the test without incident. Then, under general endotracheal anesthesia, a 5.0×19 -mm premounted balloon expandable covered stent (Jostent GraftMaster; Abbott Vascular Devices, Redwood City, Calif) was navigated over a 0.014-in microguidewire (ATW; Cordis, Johnson & Johnson, Miami Lakes, Fla) into the right cavernous ICA across the orifice of the MHT, just proximal to the origin of the ophthalmic artery. The stent graft was deployed successfully and released to a diameter of 5.3 mm when the balloon was progressively inflated to a pressure of 16 atm. An immediate control angiogram showed complete exclusion of the feeding arteries from the ICA and preservation of normal flow through the parent artery.

Next, a superselective catheterization of the right ECA feeding arteries was performed with a Marathon microcatheter over a Mirage microguidewire (ev3, Irvine, Calif). A total of 2.7 mL of Onyx-18 (ev3) was delivered into the feeding arteries using dimethyl sulfoxide preparation and the reflux-hold-reinjection technique. Complete occlusion of feeding arteries from the middle meningeal artery, as well as those from the accessory meningeal artery, artery of the foramen rotundum, and the occipital artery was achieved.

2.3. Follow-up

The final control cerebral angiogram showed complete occlusion of the DAVF and preserved patency of the right



Fig. 1. Axial T2-weighted MR images of preendovascular treatment (A) and postendovascular treatment (B) demonstrate in the preprocedure image large flow voids caused by abnormal dilatation of pontomesencephalic veins with significant mass effect and brainstem edema. The postendovascular image demonstrates high intensity in the dilated veins secondary to thrombosis and clear reduction of brainstem edema.



Fig. 2. Lateral (A) and anteroposterior (B) angiograms of the right ICA show rapid arteriovenous shunting originating from branches of the MHT. The DAVF is also fed by the middle meningeal artery, accessory meningeal artery, artery of the foramen rotundum, and occipital artery on lateral (C) and anteroposterior (D) angiograms of the right ECA. Venous drainage is primarily into right cavernous sinus with retrograde flow into left transverse-sigmoid sinus. Minimal drainage is also demonstrated via the basilar vein and the vein of Galen into the straight sinus (arrowhead), with 2 giant aneurysmal abnormal dilations in the brainstem region (arrows) on the venous-phase anteroposterior (E) and lateral (F) angiograms of ECA.

ICA (Fig. 3A and B). The patient received 75 mg clopidogrel and 100 mg aspirin daily for 6 months. The patient's neurological deficits gradually improved. At 1-month MRI follow-up, there was interval thrombosis of both varices and a 2-mm reduction in their maximal diameter (Fig. 1B). Another 1-month cerebral angiogram follow-up confirmed persistent occlusion of the fistula and patency of the right ICA, with no in-stent stenosis (Fig. 3C). The patient made an excellent clinical recovery, and he was completely neurologically intact at 18-month follow-up.

3. Discussion

Dural arteriovenous fistulae can be categorized as benign or aggressive lesions based on the presence of cortical venous reflux. Dural arteriovenous fistulae with cortical venous reflux, galenic drainage, and/or venous congestion have an aggressive presentation with subsequent bleeding, nonhemorrhagic neurological deficit, or death [20]. These aggressive DAVF subtypes can be cured with endovascular, microsurgical, and radiosurgical techniques either alone or in combination with one another [2,3,18]. Clip ligation of the principal venous outflow is a possible means to treat DAVFs and a viable alternative option to endovascular management. However, in our case, surgical ligation of the varices might decompress the pons, but it will not cure the cause of the varix, the fistula itself. In addition, radiosurgery would be a less-attractive option because of the dangerous and challenging anatomical location as well as the diffuse arterial and venous involvement. Given the significant mass effect from the giant varices, coil embolization of these abnormal structures through transvenous approach may cause flow diversion into the giant varices, worsening mass effect, and edema. We therefore chose to use a covered stent and transarterial Onyx injection to obliterate the ICA and ECA feeding arteries, respectively, thereby reducing venous congestion, mass effect, and brainstem edema.

The Onyx embolic material is a nonadhesive reliable liquid agent for endovascular treatment of cerebral and spinal arteriovenous malformations, DAVFs, and hypervascular tumors [7,11,13,15]. Siekmann et al [17] showed that transvenous placement of platinum coils and subsequent delivery of Onyx 500 could successfully occlude a transverse sinus DAVF. Of note, Onyx 500 is most commonly used in the endovascular treatment of giant aneurysms, and its suitability for many kinds of DAVFs is unknown. Suzuki et al [19] reported on a series of efficacious transvenous treatments for spontaneous dural carotid-cavernous fistulae using a combination of detachable coils and Onyx 34. Moreover, the feasibility of definitive transarterial embolization with Onyx has been recently described in certain type of intracranial DAVFs [2,3,16]. In our case, Onyx injection through multiple ECA branches was performed without any complications, and we obtained good Onyx penetration and adequate occlusion of multiple feeding arteries. The embolic material reached the shunt itself, as well as the proximal origin of the draining vein.

The use of stent grafts in the intra- and extracranial vasculature has emerged as a promising alternative to other more labor-intensive cranial endovascular therapies, such as those done for extracranial and intracranial aneurysms, carotid-cavernous fistulae, and pseudoaneurysms [1,4,6,10,12,14]. Saatci et al [14] reported a midterm follow-up on 24 patients with ICA aneurysms who were successfully treated by using a Jostent coronary covered stent, which was deployed in the parent artery to cross the aneurysm neck. All but 4 of these aneurysms were extradural, and they were located in the petrous or cavernous ICA. Twenty-three aneurysms were immediately excluded from the circulation after stent placement. All mass effect symptoms were resolved during the follow-up period [14].



Fig. 3. The immediate postprocedure lateral angiograms with right ICA (A) and ECA (B) injection show complete obliteration of the DAVF and preservation of right ICA. One month later, angiography confirms no recanalization of the fistula with patency of the right ICA on the angiogram of the right common carotid artery (C).

The main technical limitation of stent grafts is their restricted flexibility and high rigidity, which hinders navigation in the curvaceous cerebral vasculature [9]. The placement of covered stents may produce complications including dissection and vasospasm of the cerebral arteries. Cases with significant tortuousity are not suitable candidates for currently available covered stents. Of further concern is the potential risk of exclusion of side or perforating branches, originating from the treated cerebral arterial segment, and which may result in ischemic complications, notably in deep areas with limited potential collateral supply [1,14]. Another potential concern is delayed in-stent stenosis after stent placement. An experimental aneurysm study showed a tendency for covered stents to develop progressive neointimal proliferation, which may result in stenosis of 60% or more. Nonetheless, the degree of observed stenosis within this experimental study was not hemodynamically significant, and the lack of subsequent clopidogrel or other antiplatelet agent administration may have contributed to delayed stenosis [8]. In-stent stenosis has also been observed in a small number of patients after the Neuroform stent placement for cerebral aneurysm treatment. Although the most of these delayed stenosis are asymptomatic and may spontaneously resolve on further follow-up, symptomatic patients may require angioplasty or surgical bypass [5]. It is therefore necessary to perform long-term follow-up to exclude any delayed in-stent stenosis despite complete fistula occlusion and lack of evidence for ICA stenosis at this patient's 1-month cerebral angiogram.

Despite these limitations, we believe that a variety of neuropathologic entities, particularly those involving the cavernous sinus region, such as dural fistulae, cavernous aneurysms, and tumors of the skull base, may be amenable to covered stent placement for the occlusion of branches or aneurysms originating from the cavernous ICA while preserving the ICA lumen.

4. Conclusion

The combined technique of placement of a covered stent and transarterial Onyx delivery can be very efficacious in treating complex DAVFs, which may be supplied by both ECA and cavernous ICA branches. The excellent clinical and angiographic results in this patient should encourage further study to evaluate the effectiveness and safety of intracranial placement of covered stent. By engineering more flexible and low-profile covered stents, we may be better able to circumnavigate the intracranial vasculature, and this will add another powerful tool to the endovascular armamentarium for treating intracranial vascular diseases.

Acknowledgments

We thank Fernando Viñuela, MD, Division of Interventional Neuroradiology, University of California at Los Angeles Medical Center, Los Angeles, Calif, for advice and editorial assistance in the preparation of this article.

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