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Transarterial Onyx Embolization for Patients with Cavernous Sinus Dural Arteriovenous Fistulas Who Have Failed Transvenous Embolization

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Abstract Transvenous embolization is the treatment of choice for cavernous sinus dural arteriovenous fistulas (csDAVFs) despite occasional difficulty in transvenous catheterization. We reported our experience in the treatment of csDAVFs by transarterial Onyx embolization in patients who had failed transvenous catheterization. We reviewed the clinical and radiographic records of csDAVFs patients receiving transarterial Onyx embolization after failed transvenous Onyx embolization at our institution over a period of 31 months. Success was defined as complete or near complete occlusion upon angiographic examination. In seven cases, the microcatheter failed to reach the cavernous sinus; in the remaining case, the internal jugular vein was occlusive. Eight sessions of the embolization and catheterization procedures via the arterial routes were conducted. Among them, five cases via the middle meningeal artery and the other three via the accessory meningeal artery. Angiography, immediately after embolization, revealed complete occlusion in seven cases (87.5 %) and partial occlusion in the remaining case. Angiographic follow-up (range, 6–10 months) showed that all patients achieved complete embolization. In cases where transvenous embolization of the cavernous sinus is difficult, transarterial embolization of the fistulas offers a safe and effective alternative.

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Introduction

Intracranial dural arteriovenous fistulas (DAVFs) account for 10–15 % of all intracranial arteriovenous malformations [1]. The cavernous sinus is a region where fistulas frequently occur and, in most cases, cavernous sinus DAVFs (csDAVFs) involve the bilateral cavernous sinuses. Cavernous sinus DAVFs typically have multiple feeding arteries originating from the internal carotid artery (ICA) and/or external carotid artery (ECA).

Patients with csDAVFs may develop such alarming symptoms as proptosis, chemosis, extraocular muscle palsies, and threatened visual loss due to increased intraocular pressure or reduced ocular perfusion pressure, requiring rapid curative treatment. Endovascular therapy has become the main therapeutic modality for symptomatic csDAVFs [2]. Currently, transvenous embolization of the cavernous sinus is the preferred choice. However, catheterization through the conventional route, such as the inferior petrosal sinus or superior ophthalmic vein [3–5], occasionally fails to reach the cavernous sinus [6]. Surgical cannulation of the superior ophthalmic vein or percutaneous transorbital extraconic puncture of the cavernous sinus for catheterization has been proposed, but carries the risk of a number of complications, such as orbital hematoma or infection and damage to adjacent nerves [7]. Approach via cortical veins such as the superficial temporal veins [8] and superficial middle cerebral veins [9] to catheterize the cavernous sinus has been attempted; however, exposure of these cortical veins entails craniotomy and complicated procedures.

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Transarterial embolization of csDAVFs with liquid embolic agents has a higher risk of embolization of dangerous anastomoses between the dural branches of the ECA and the ICA, the vasa nervorum, ophthalmic artery, or vertebral artery [6, 7]. Onyx (ev3, Plymouth, MN, USA) has become the preferred embolysate due to its cohesive and non-adhesive nature that assists in controlled penetration of the fistula [10, 11]. We think this merit can reduce the risk. Recently, Zhang et al. reported a satisfactory outcome of transarterial Onyx embolization in eight patients with csDAVFs with a mean follow-up of 21.6 months [12]. Pero et al. reported the treatment of csDAVFs by Onyx injection through the ascending pharyngeal artery in two patients who had failed the transvenous approach through the inferior petrosal sinus or superior ophthalmic vein [6]. Gandhi et al. reported a case of a csDAVFs that was successfully treated with transarterial embolization with Onyx through the distal internal maxillary artery after multiple failed attempts to catheterize the cavernous sinus [13]. Amiridze et al. also described a case of csDAVFs that was cured with Onyx embolized through middle meningeal artery and sphenopalatine artery [14]. All of them lacked summary of treatment experience. In the current paper, we report our experience and the outcome of Onyx embolization via the arterial route for eight patients with csDAVFs who failed transvenous embolization.

Materials and Methods

We reviewed the full clinical records, angiograms, and procedure reports of patients with csDAVFs who had failed transvenous Onyx embolization and received transarterial Onyx embolization instead at our department between November, 2010 and June, 2013. The study protocol was approved by the local institutional review board at the authors' affiliated institution. All patients provided consent to the embolization procedure. Consent for the current study was not required because of its retrospective nature.

All patients received ophthalmological examination by an ophthalmologist before treatment to obtain baseline data for visual acuity and fields for comparison with subsequent studies. Conventional cerebral angiography was performed under intravenous general anesthesia via the femoral artery (Artis BA, Siemens, Erlangen, Germany). Heparin was injected to keep the activated clotting time of the patients between 200 and 300 s. Angiography included bilateral selective ICA and ECA angiography and vertebral artery angiography to identify all arterial supplies to the fistulae. All imaging results were reviewed by two interventional neuroradiologists, and fistulous site and arterial feeders,

venous drainage patterns, collateral flows and dangerous vascular anastomosis were then evaluated before management decisions were made. DAVFs were categorized according to both the Barrow classification [15] and the Borden-Shucart classification system [16]. The final decision of treatment strategies was made after careful analysis of the clinical and imaging findings in each patient. When patients who had failed transvenous embolization through superior and inferior petrosal sinus, the arterial route was considered.

Transarterial embolization was performed in patients who had failed the transvenous embolization procedure by positioning a 6F Envoy guiding catheter (Cordis, Hialeah, FL) in the ECA. The contralateral femoral artery was catheterized for control angiography. A microcatheter (Echelon series, Micro Therapeutics or Marathon flow-directed catheter, ev3, Plymouth, MN, USA) was navigated over a microguidewire (Transend series, Boston Scientific, Boston, USA; or Mirage 008, ev3, Plymouth, MN, USA) to reach the distal aspect of the arterial pedicle that fed the fistula. Super-selective angiography was then performed to confirm optimal wedging of the microcatheter and to identify the normal arterial branches and dangerous anastomoses.

Onyx was slowly injected using the "reflux–hold–reinjection" technique under the real-time roadmap. When reflux was observed to flow into the non-targeted area, the infusion was held for 20 s to 2 min to allow the Onyx to precipitate and form a plug around the catheter. Reinjection was carried out slowly in a controlled manner, engendering a new road map. Each time, the real-time roadmap was refreshed to prevent confusion on the progression of Onyx. Angiographic images of all previously known fistulous feeders were used to evaluate the residual DAVFs, whether the normal branches and dangerous anastomoses were embolized or not. Heparin injection was discontinued but not reversed at the end of the procedure. Routine postoperative care was given to all patients in the Neurosurgical Intensive Care Unit.

All patients underwent CT scan immediately after the procedure, and a neurologic examination was performed in all cases at discharge. Angiographic follow-up was performed in all patients. The evaluation criteria for angiographic results were defined as total occlusion of the shunt. This was defined as non-recognizable arteriovenous shunts and complete occlusion of the draining vein, near total occlusion, which was defined as a small residual stagnant shunt without cortical reflux or ophthalmic venous drainage, and partial occlusion, which manifested as only flow reduction with clear residual shunt. Complete and near complete occlusions were considered successful angiographic results [17, 18].

Results

Eight (17.8 %, 8/45) cases failed transvenous Onyx embolization and received transarterial Onyx embolization instead. Transvenous embolization was abandoned because the microcatheter failed to reach the cavernous sinus in seven patients and because the internal jugular vein below the jugular bulb was occlusive in another patient. They included four males and four females with a median age of 36 (range, 26–57) years. Four patients had Barrow type C fistula, and four patients had Barrow type D fistula. All patients had Borden type I DAVF. Five (62.5 %, 5/8) cases involved the left cavernous sinus and three (37.5 %, 3/8) cases involved the right cavernous sinus. None had received prior medical therapy before the failed transvenous embolization. All patients (100 %, 8/8) had conjunctival congestion and five (62.5 %, 5/8) patients had proptosis and chemosis. The presenting symptoms of the patients are shown in Table 1.

In all, eight sessions of embolization and catheterization procedures through the arterial routes were conducted: five via the middle meningeal artery and three via the accessory meningeal artery. One (12.5 %, 1/8) patient exhibited no symptoms and seven (87.5 %, 7/8) patients demonstrated marked symptom improvement at discharge. All patients were discharged at 3–8 (median, 5.5) days after the embolization. The patients were followed up for a median duration of 14.5 (range, 6–26) months. Six (75 %, 6/8)

patients became asymptomatic and two (25 %, 2/8) further improved at the final follow-up visit (Table 2).

Angiography taken immediately after embolization revealed that seven (87.5 %, 7/8) patients achieved total occlusion, and one (12.5 %, 1/8) patient had partial occlusion. The patients were followed up by angiography for a median duration of 6.0 (range, 6–10) months. All patients achieved complete embolization at the final follow-up.

Complications related to transarterial embolization procedures occurred in two (25 %, 2/8) patients, including left ipsilateral facial numbness in one case and right abducens paralysis and chemosis which were contralateral to the embolic approach in the other patient. Both complications were cured by intravenous methylprednisolone at a daily dose of 120 mg for a total of 3 days and mannitol at a dose of 50 g, divided into two equal doses and given every 12 h, for a total of 7 days.

Case 1

A 26-year-old woman presented with headache, diplopia, and left conjunctival congestion for 3 months. There was no history of cranial trauma and treatment. Angiography was performed for definitive diagnosis and therapeutic planning. Arteriography showed arteriovenous shunts in the dura mater on the left of the cavernous sinus, which was supplied mainly by branches of the middle meningeal artery, and draining exclusively into the left inferior petrosal sinus. We attempted to catheterize to the left cavernous sinus through the inferior petrosal sinus, but failed. We chose the middle meningeal artery as the route to

Table 1 The presenting symptoms of the patients

Characteristics	N(%)
All patients	8 (100)
Age, years	
Median	36
Range	26,57
Male sex	4 (50)
Symptoms	
Conjunctival congestion	8 (100)
Chemosis	5 (62.5)
Proptosis	5 (62.5)
Headache	3 (37.5)
Periorbital bruit	2 (25)
Diplopia	1 (12.5)
Visual impairment	1 (12.5)
Borden type	
I	8 (100)
Barrow type	
C	4 (50)
D	4 (50)

Table 2 Outcome of eight patients with cavernous sinus dural arteriovenous fistulas

Outcome	N (%)
Clinical outcome	
Discharge	
Symptom free	1 (12.5)
Improvement	7 (87.5)
Follow-up	
Symptom free	6 (75)
Further improvement	2 (25)
Angiographic outcome	
Immediate postoperation	
Total occlusion	7 (87.5)
Partial occlusion	1 (12.5)
Follow-up	
Total occlusion	8 (100)
Recurrence	0 (0)

embolize the fistulas with 1.5 volume Onyx. We achieved immediate total occlusion, which was further observed at the follow-up angiography (Fig. 1).

Case 2

A 30-year-old woman presented with proptosis, chemosis, conjunctival congestion, and blurred vision of the left eye for 5 months. There was no history of cranial trauma and treatment. Angiography was performed for definitive diagnosis and therapeutic planning. The arteriography showed arteriovenous shunts in the dura mater on the left of the cavernous sinus, supplied mainly by branches of the accessory meningeal artery, and draining exclusively into the left SOV. After transvenous embolization of the cavernous sinus, we chose the accessory meningeal artery as the route to embolize the fistulas with 1.5 volume Onyx. Total occlusion was achieved immediately and at the follow-up angiography (Fig. 2).

Discussion

The natural course of csDAVFs is more benign than that of DAVFs at other sites. Reportedly, spontaneous regression occurs in 10–73 % of the patients [19, 20]. However, once

csDVAF becomes symptomatic, curative treatment should be promptly given in order to prevent visual deterioration [21]. In our series, the patients exhibited severe conjunctival congestion, apparent proptosis and chemosis, intolerable diplopia, persistent headache or even vision lesion, necessitating urgent treatment. Our therapeutic strategy is based on a comprehensive review of such factors as the feeding arteries, venous drainage, location of the fistulas, clinical manifestations, baseline imaging studies, patient condition and social factors [2]. Transvenous embolization is considered an effective and safe option for csDAVFs [22, 23], which is usually performed via the inferior petrosal sinus or superior ophthalmic vein. Nevertheless, percutaneous catheterization through the inferior petrosal sinus and superior ophthalmic vein fails occasionally, mandating the use of alternative access routes that are acceptable and feasible. In our another study, for nine patients who had failed embolization via the inferior petrosal sinus, we resorted to surgical cannulation of the superior ophthalmic vein for catheterization, a method reported by other investigators [24, 25]. Some researchers used the cortical veins as the surgical route to catheterize the cavernous sinus [8, 9]. We think that these methods may incur considerable risk of injuries to adjacent important structures and involve complicated procedures. Our preliminary experience led us to believe that, if Onyx can be effectively
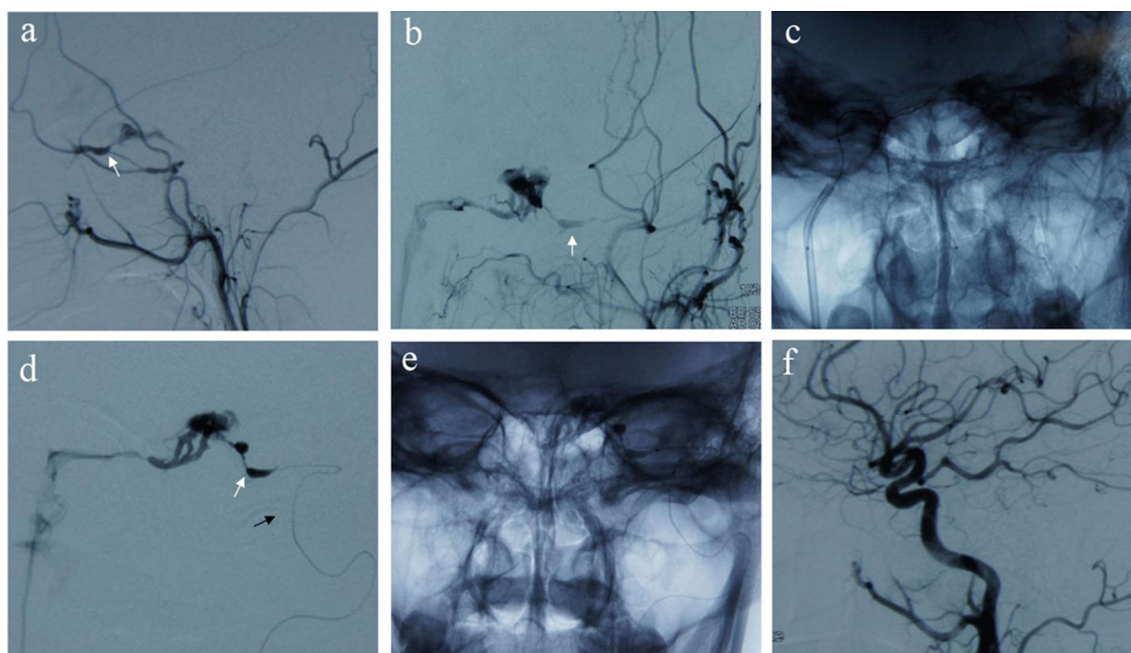
Fig. 1 Digital subtraction angiography in a 54-year-old woman with headache, diplopia, and *left* conjunctival congestion for 3 months shows embolization of the cavernous dural arteriovenous fistula via the middle meningeal artery. **a**, **b** Arteriography of the left ECA shows dural arteriovenous fistula (*white arrow*), the contralateral inferior petrosal sinus and intercavernous sinuses. **c** A microcatheter was advanced but failed to catheterize the cavernous sinus via the contralateral inferior petrosal sinus. **d**,**e** The microcatheter was guided through the *left* middle meningeal artery (*black arrow*) and cast of Onyx was injected (*white arrow*). **f** Arteriography of the *left* carotid artery shows complete occlusion
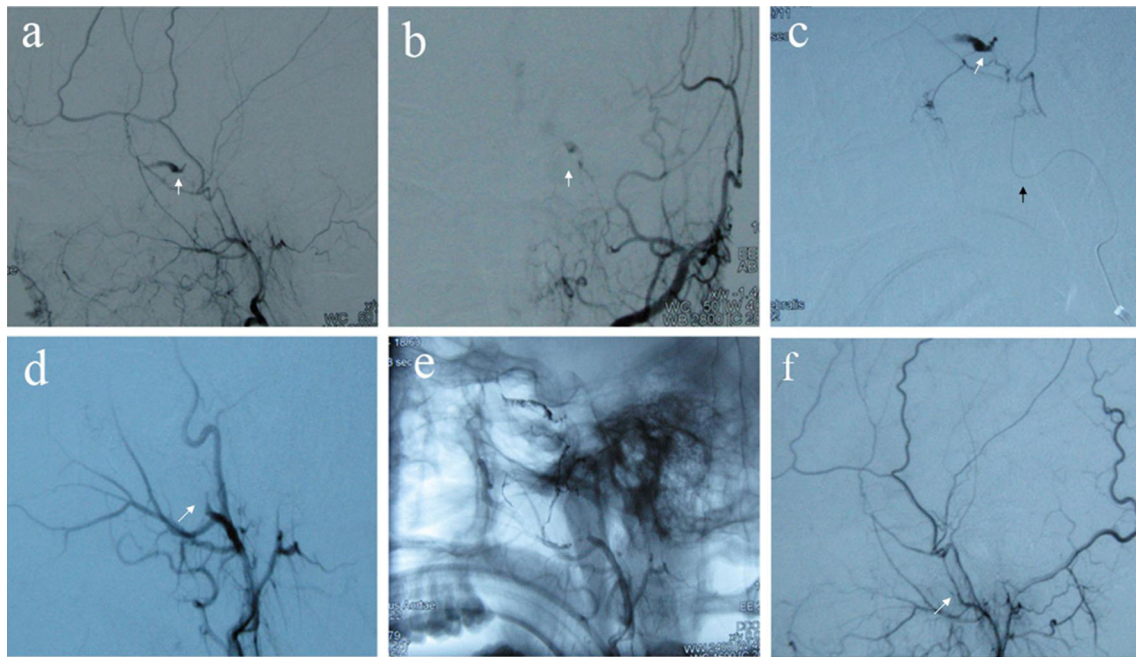
Fig. 2 Digital subtraction angiography in a 54-year-old woman with proptosis, chemosis, conjunctival congestion, and blurred vision of *left* eye for 5 months shows embolization of the cavernous dural arteriovenous fistula via the anterior meningeal artery. **a, b** Arteriography of the *left* ECA shows dural arteriovenous fistula (*white arrow*). **c** A microcatheter was advanced to catheterize to the *left* accessory meningeal artery (*black arrow*) and super-selective angiography was carried out. **d** The cast of Onyx (*black arrow*). **e, f, g** Arteriography of the *left* ECA shows complete occlusion immediately following embolization, with apparent occlusion of the middle meningeal artery. **h** Arteriography of the *left* ECA shows complete occlusion at the 6 month follow-up, the middle meningeal artery is developed again

applied and embolization of dangerous anastomoses avoided, the arterial route provides an acceptable, viable alternative route and is worth considering. Moreover, this method can maintain the patency of the cavernous sinus, which is more in conformity with the physiology.

According to our experience, using the "reflux-hold-reinjection" technique, Onyx can migrate in a retrograde fashion through a single or several pedicles into the arterial feeders from other vessels, and achieve deeper penetration and occlusion of the entire fistulous network. Additionally, Onyx has better penetration quality, less adhesiveness, but cohesiveness, and slow polymerization, which allows a prolonged injection and better controllability [10, 11]. These merits enable a large volume of Onyx to be delivered via a single or a few injections, a circumstance that would make feasible successful transarterial embolization of csDAVFs. Avoiding embolization of dangerous anastomoses remains a critical issue in transarterial embolization.

In our series, eight patients were embolized via the arterial route. The internal jugular vein below the jugular bulb was occlusive in one patient as was displayed by cerebrovascular imaging. Catheterization in seven patients was conducted through the inferior and superior petrosal sinus, but failed. We found that all the fistulas were limited in size with few feeding arteries and slow blood flow. Furthermore, the sites of fistulas were further away from the cavernous sinus. These considerations prompted us to pursue transarterial embolization for the eight patients.

According to Rossitti, the dural vessels allow larger amounts of reflux plugs as long as dangerous anastomoses are avoided when compared to other vessels [26]. The middle meningeal artery provides an excellent route for the delivery of Onyx to occlude the fistula and the retrograde artery to the fistula, thereby occluding arterial feeders from the branches of the ECA or the ICA. Some authors reported their experience in using the middle meningeal artery as a conduit for Onyx embolization, and achieved high cure rate [27–29]. Hu et al. used the route and achieved an angiographic cure rate of 77.1 % (27/35) [29]. But these reports mainly describe non-csDAVFs. There is a case of csDAVFs that was embolized through middle meningeal artery with Onyx, but the case experienced a rapid decrease in heart rate followed by asystole which was reported in the literature [14]. We applied the technique in treating csDAVFs by using the middle meningeal artery as a single pedicle for embolization in five cases of csDAVFs, and all achieved total occlusion at the immediate and follow-up angiography. One patient showed chemosis and abducent nerve palsy in the contralateral eye, as Gandhi et al. reported [13], which the reason may be an inflammatory response generated by Onyx. None occurred hemodynamic instability, such as

bradycardia or asystole. Nevertheless, close attention should be paid to potential risks. The anterior group of the middle meningeal artery occasionally gives a branch to anastomose with the ophthalmic artery through the superior orbital fissure, and the medial branch of the skull base group of the middle meningeal artery supplies the trigeminal ganglion. This may explain the reason of hemodynamic instability of the case that was embolized through middle meningeal artery with Onyx [14].

The accessory meningeal artery often participates in supplying csDAVFs. We have tried to use the accessory meningeal artery as a single pedicle to embolize csDAVFs in three cases. All achieved total occlusion at the immediate and follow-up angiography. But one case showed left facial numbness ipsilateral to the lesion. We thought that the complication may result from the blockage of the superior branch of the accessory meningeal artery because the branch goes through the foramen oval to partially supply the trigeminal ganglion. Additionally, there is an anastomosis between the accessory meningeal artery and the inferolateral trunk of the cavernous sinus of the ICA [30].

Two patients showed complications associated with embolization of the fistulas: one associated with dangerous anastomoses and the other may be an inflammatory response generated by Onyx. How to avoid the embolization of dangerous anastomoses remains a critical issue in transarterial embolization. Super-selective angiography should be done before Onyx is injected as it may help to identify dangerous anastomoses, thereby facilitating confirmation of optimal wedging of the microcatheter and refluent distance of Onyx. Super-selective angiography is not a very reliable method to detect anastomoses and in this regard anatomical knowledge is very important. Sometimes, in order to avoid unwanted infusion of Onyx into potentially dangerous anastomoses, we can also place a hyperglide balloon in the ICA where dangerous anastomoses exist to protect the ICA. The same approach can be applied to the meningeal artery so that the important vessels can be protected. Hyperglide balloon placement can also prevent Onyx reflux and enable injection of Onyx into more feeding vessels [12].

In our center, although transvenous embolization is the preferred approach, when catheterization through the venous route fails, we think that transarterial embolization offers a feasible and effective approach to treat csDAVFs if the fistulas involve the dura mater of the cavernous sinus of only one side and are limited in size with few feeding arteries and the availability of a proper arterial route. The middle meningeal artery and the accessory meningeal artery may be utilized for transarterial embolization. Recently, Pero et al. have reported three cases of csDAVFs treated by Onyx embolization through the superior pharyngeal branch of the ascending pharyngeal artery [6].

However, our series so far contains the largest number of patients who had failed transvenous embolization of csDAVFs and treated by transarterial Onyx embolization instead. We further focused on discussing the merits and risk of the middle meningeal artery and the accessory meningeal artery acting as the transarterial routes to embolize csDAVFs. However, the number of our cases is still small, and we will continue to collect data and summarize our experience.

Conclusions

When transvenous embolization of the cavernous sinus to treat csDAVFs encounters difficulty, transarterial embolization of the fistulas offers a safe and effective alternative. Furthermore, the middle meningeal artery and the accessory meningeal artery can provide good transarterial routes for embolization of csDAVFs.

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