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# Treatment of cavernous sinus dural arteriovenous fistula using different surgical approaches: Analysis of 32 consecutive cases



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| ARTICLE INFO  | A B S T R A C T  |
|---|--|
| <i>Keywords:</i><br>Cavernous sinus<br>Dural arteriovenous fistula<br>Embolization<br>Intravascular treatment | Objective: Transarterial and transvenous embolization methods are considered effective and safe approaches for<br>the treatment of cavernous sinus dural arteriovenous fistula (CSDAVF). Here., we report the angioarchitectural<br>features and clinical outcomes of CSDAVF in patients treated with either the inferior arterial approach (IAA) or<br>the inferior petrosal sinus approach (IPSA).<br><i>Methods</i> : The clinical data of 32 patients with CSDAVF treated at our institution from May 2008 to May 2014 were<br>retrospectively analyzed. All patients underwent routine diagnostic digital subtraction angiography (DSA) before<br>surgery. Embolization was performed using the IPSA through the internal jugular vein or IAA, based on<br>angioarchitectural features.<br><i>Results</i> : Of the 32 patients with CSDAVF, 24 underwent embolization treatment through the internal jugular vein-<br>IPSA and 8 patients underwent treatment through IAA. Nineteen patients in the IPSA group experienced mild<br>headache, which improved after specific treatment. The immediate postembolization angiographic results<br>revealed complete occlusion in 26 cases (18 IPSA and 8 IAA) and almost complete occlusion in 6 cases (IPSA).<br>Complications that occurred during the procedure included abducens nerve palsy (n = 1, IPSA) and prosopoplegia<br>(n = 1, IAA). One patient developed tinnitus, which was diagnosed as anterior cranial fossa new-onset dural<br>arteriovenous fistula on DSA, whereas the symptoms of other patients all improved with no recurrence.<br><i>Conclusions</i> : On the basis of the angioarchitectural features of CSDAVF, IAA can be considered the primary<br>treatment when the blood-supplying artery and fistula are relatively singular, and when the microcatheter can<br>easily reach the fistula through the artery. The venous approach should be selected as the primary approach when<br>the fistula is indistinguishable and blood is supplied by multiple arteries through small plexiform vessels.<br>Choosing the optimal surgical approach may increase the success rate of intravascular CSDAVF surgery and may<br>help a |

# Introduction

Cavernous sinus dural arteriovenous fistula (CSDAVF) is characterized by abnormal shunts between the arterial and venous circulation developing on the dura mater, usually within or near the walls of a dural sinus.<sup>8,21</sup> These fistulas arise spontaneously and represent approximately 10–15% of all intracranial arteriovenous shunts.<sup>11</sup> Patients with CSDAVF may develop signs of orbital hypertension, including glaucoma, pulsatile exophthalmos, chemosis, conjunctival injection, and episcleral venous dilation.<sup>14,20</sup> However, the symptoms of CSDAVF depend on the location of the shunt, type of venous drainage, and flow characteristics.<sup>5</sup> Owing to the special anatomical features of the cavernous sinus, CSDAVFs have various clinical manifestations and thus may necessitate various treatment approaches. CSDAVFs are usually managed with carotid compression, endovascular intervention, surgery, radiosurgery, or a combination of multidisciplinary approaches.<sup>7,13,24</sup> However, endovascular techniques have become the most common treatment for patients with symptomatic CSDAVFs.<sup>6</sup> Both the transvenous and transarterial approaches of endovascular embolization are widely accepted and considered effective and safe options for the treatment of CSDAVFs.<sup>2</sup>

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Although recent studies have shown good outcome with low complication rates for occluding dural arteriovenous fistulas (DAVFs) via the transarterial approach, this route may not always be feasible, especially if multiple feeders are present and shunt into a widely dispersed segment of the dural wall.<sup>5</sup> In such conditions, transvenous approaches may be considered. Among different transvenous routes, the inferior petrosal sinus approach (IPSA) is the most preferred and feasible route, even when the sinus is partially or completely thrombosed.<sup>1</sup> In this study, we retrospectively analyzed the angioarchitectural features of CSDAVF including the operative techniques and treatment outcomes based on different surgical procedures.

# Methods

# Patient population

In this retrospective study, a total of 32 patients with CSDAVFs treated at our institution from May 2008 to May 2014 were included in the final analysis. The patient population included 7 male and 25 female patients, aged from 5 to 80 years (average, 50.8 years). Signs and symptoms included history of previous craniocerebral trauma in 8 patients, ocular hyperemia in all patients, combined mild proptosis in 19 patients, diplopia in 6 patients, vision loss in 8 patients, intracranial murmur in 14 patients, and history of previous repeated nasal bleeding in 2 patients. The disease course, defined as the time from when the patient reported the presence of symptoms to the time of surgery, ranged from 2 to 18 months (average, 3.5 months). The manifestations on digital subtraction angiography (DSA) were classified according to Barrow types,<sup>1</sup> resulting in 15 cases of type C simple external carotid artery dural shunts and 17 cases of dural shunts from the meningeal branches of the external and internal carotid arteries. All patients had ophthalmic venous drainage, including 18 cases with combined inferior petrosal sinus drainage, 7 with combined cortical venous drainage, 4 with superior petrosal sinus drainage, 2 with combined pterygoid plexus drainage, and 3 with combined basal vein drainage.

# Treatment methods

All patients underwent routine diagnostic DSA before surgery. Embolization was performed through internal jugular vein-IPSA or the inferior arterial approach (IAA) based on angioarchitectural features. The details of the procedures are described below. The surgical approach was chosen mainly based on three factors: (i) blood supply, (ii) position of the DAVF (front, simple blood-supplying-artery approach; back, vein approach), and (iii) draining vein.

#### Internal jugular vein-inferior petrosal sinus approach

Routine left femoral artery and right femoral vein puncture was performed, and 5F and 6F artery sheaths were implanted. A 4F DSA catheter was connected to a high-pressure drip then guided into the main blood-supplying artery for contrast agent deposition. By using a coaxial technique, a 120-cm 4F catheter was inserted inside a 6F guiding catheter through the femoral vein and placed close to the internal jugular vein bulb on the lesioned side. A 0.035-inch loach guidewire was used to enter the inferior petrosal sinus and access the petrous apex, and the 4F catheter was then placed into the inferior petrosal sinus. An Echelon<sup>™</sup>-14 microcatheter (Medtronic, USA) was inserted into the inferior petrosal sinus directly through the 120-cm 4F catheter with the aid of a Traxcess-14 microguidewire (Microvention, USA), and the tip was placed into the dilated ophthalmic superior/inferior vein. Once the location was confirmed using super-selective angiography, the surgery was terminated with filling of coils until the fistula was completely embolized or the draining vein showed obvious slowing.

#### Inferior arterial approach

The right femoral artery was punctured, a 5/6F artery sheath was implanted, and the proper blood-supplying artery was chosen based on the diameter, length, tortuosity, and type of collateral branch determined using super-selective angiography. An Echelon<sup>™</sup>-10 or Marathon<sup>™</sup> microcatheter (Medtronic, USA) was used to super-selectively approach the cavernous sinus fistula with the aid of the microguidewire, and the angioarchitecture was revealed through super-selective angiography, thus avoiding dangerous anastomoses. Spring coil embolization was performed on the fistula to slow down the blood flow, when necessary. Subsequently, Onyx<sup>™</sup> LES (Medtronic, USA) was slowly injected using the roadmap-bolus-diffusion-pause-roadmap-bolus method. Angiography was repetitively conducted during the Onyx injection to observe the status of the cavernous sinus embolization. The surgery was terminated once the fistula disappeared.

# Results

Of the 32 patients with CSDAVF, 24 underwent embolization treatment through internal jugular vein-IPSA and 8 patients underwent treatment through IAA. Among the IPSA cases, one failed case was successfully treated with Onyx after switching to IAA and another failed case was treated by puncturing the superior ophthalmic vein (Fig. 1). Nineteen patients in the IPSA group experienced mild headache symptoms after surgery, which improved after specific treatments. One patient showed postoperative abducens nerve palsy, which improved after 2 months. The clinical characteristics of the study population are presented in Table 1 (see Fig. 2).

Among eight IAA cases, two cases were failures and were successfully embolized through IPSA. Six cases were treated with Onyx embolization alone (including one case that failed with the venous approach and was switched to IAA), and one was treated with spring coils combined with Onyx embolization.

Angiography conducted immediately after embolization showed complete fistula occlusion in 26 cases (18 IPSA and 8 IAA) and almost complete occlusion in 6 cases (IPSA). All ocular symptoms improved or healed after embolization, and all intracranial murmurs disappeared. Complications that occurred during the procedure included abducens nerve palsy (n = 1, IPSA) and prosopoplegia (n = 1, IAA). The average duration of the follow-up of patients was 28 months; 27 patients completed the follow-up, whereas 5 were lost to follow-up. One patient showed tinnitus, which was diagnosed as anterior cranial fossa newonset DAVF on DSA, whereas the symptoms of other patients all improved with no recurrence (Table 2).

#### Illustrative case

A 50-year-old female patient was hospitalized after 3 months for hyperemia and swelling of the left eye. DSA showed DAVF in the left cavernous sinus region: only the left external carotid artery branches were responsible for main blood supply; the fistula was located in the gap in front of the cavernous sinus region, with blood reflux through the ophthalmic vein (Fig. 1). After the embolization procedure, DSA showed complete occlusion of the DAVF. The congestion symptom disappeared after surgery. The patient's clinical status improved, and there were no postoperative complications and recurrence at the 6-month follow-up.

# Discussion

Currently, the treatment plans for CSDAVF mainly include intravascular interventional surgery, neck compression, and gamma knife treatment. As reported in previous studies,<sup>3,18,22,25</sup> the healing rate of CSDAVF with interventional embolization is between 66% and 89%, with significant treatment efficacy, and embolization through the venous approach is usually considered the primary, safe, and effective



**Fig. 1.** Study design. Eligible patients were assigned to undergo an embolization procedure through the internal jugular vein-inferior petrosal sinus approach (IPSA) or the inferior arterial approach (IAA). In the IPSA group, one patient was switched to IAA and one patient was switched to the eye vein approach (EVA). In the IAA group, two patients were switched to IPSA. The average duration of the follow-up of patients was 28 months. A total of 27 patients completed the follow-up and 5 were lost to follow-up. CSDAVF, cavernous sinus dural arteriovenous fistula.

Fig. 2. A female patient, age 50 years, had been experiencing hyperemia and swelling of the left eye for > 3 months. Digital subtraction angiography (DSA) showed a dural arteriovenous fistula in the left cavernous sinus region: only the left external carotid artery branches are responsible for main blood supply; the fistula is located in the gap in front of the cavernous sinus region, with blood reflux through the ophthalmic vein. 1. The external carotid artery branches responsible for blood supply are shown by white arrows; the fistula location is shown by the circle; and the dilated superior ophthalmic vein is shown by black arrows. 2. With the left internal jugular vein-inferior petrosal sinus approach, the microcatheter could not reach the gap where the fistula is located after reaching the cavernous sinus region. 3. After the external carotid artery approach, the microcatheter could reach the fistula (circle) through middle meningeal artery. 4. After injecting 0.1 mL of Onyx (the arrow indicates the cast). 5. Postoperative DSA showing that the fistula has disappeared.





method.<sup>12,23</sup> However, considering the angioarchitectural features of individual patients with CSDAVF, the arterial approach can sometimes also be considered as the primary treatment plan.

In this study, 24 patients underwent embolization treatment through internal jugular vein-IPSA for the first time. Among them, 13 showed good angiography results for the inferior petrosal sinus with draining to the jugular venous bulb, whereas 11 did not undergo imaging of the inferior petrosal sinus. In one case, the arterial approach failed and was switched to internal jugular vein-IPSA; in the end, the microcatheter was successfully guided into the cavernous sinus through the inferior petrosal

#### Table 1

Baseline characteristics of the study population.

| Characteristics                           | N = 32     |  |
|---|------------|--|
| Age (years)                               | 50.80      |  |
| Sex, n (%)                                |            |  |
| Male                                      | 07 (21.9)  |  |
| Female                                    | 25 (78.1)  |  |
| Initial presenting symptoms, n (%)        |            |  |
| Craniocerebral trauma                     | 0 8 (25.0) |  |
| Ocular hyperemia                          | 32 (100)   |  |
| Mild proptosis                            | 19 (59.4)  |  |
| Diplopia                                  | 06 (18.8)  |  |
| Vision loss                               | 08 (25.0)  |  |
| Intracranial murmur                       | 14 (43.8)  |  |
| Nasal bleeding                            | 02 (6.25)  |  |
| Embolization technique, n (%)             |            |  |
| Inferior petrosal sinus approach          | 24 (75.0)  |  |
| Inferior arterial approach                | 08 (25.0)  |  |
| Angiographic classification, n (%)        |            |  |
| Type C                                    | 15 (46.9)  |  |
| Type D                                    | 17 (53.1)  |  |
| Drainage patterns, n (%)                  |            |  |
| Ophthalmic venous drainage                | 32 (100)   |  |
| Combined inferior petrosal sinus drainage | 18 (56.2)  |  |
| Combined cortical venous drainage         | 07 (21.9)  |  |
| Superior petrosal sinus drainage          | 04 (12.5)  |  |
| Combined pterygoid plexus drainage        | 02 (6.25)  |  |
| Combined basal vein drainage              | 03 (9.4)   |  |

# Table 2

Postoperative results of the inferior petrosal and inferior arterial approaches.

| Characteristics   | (N = 32)          |  |  |
|---|-------------------|--|--|
| Procedural complications, n (%)                             |                   |  |  |
| Abducens nerve palsy  | 1 (3)             |  |  |
| Prosopoplegia   | 1 (3)             |  |  |
| None  | 30 (94)           |  |  |
| Angiographic results immediately after the procedure, n (%) |                   |  |  |
| Complete  | 26 (81)           |  |  |
| Almost complete   | 6 (19)            |  |  |
| Incomplete  | 0 (0)             |  |  |
| Patients who completed follow-up, n (%)                     | 27 (84)           |  |  |
| Including angiography                                       | 9                 |  |  |
| By phone call   | 18                |  |  |
| Average clinical follow-up, months                          | $28^{a} (n = 27)$ |  |  |
| Angiographic results at follow-up                           | (n = 9)           |  |  |
| Recurrence  | 1 <sup>b</sup>    |  |  |
| No recurrence   | 8                 |  |  |
| Symptoms at follow-up                                       | (n = 27)          |  |  |
| Ocular  | 0                 |  |  |
| Intracranial murmur   | 0                 |  |  |
| Tinnitus  | 1                 |  |  |
| None  | 26                |  |  |
| Clinical outcome at follow-up                               | (n = 27)          |  |  |
| Improved  | 26                |  |  |
| Mild residue  | 1                 |  |  |
| Not improved  | 0                 |  |  |

<sup>a</sup> Angiographic follow-up was conducted at approximately 6 months, and follow-up via phone call ranged from 3 to 62 months.

<sup>b</sup> One patient developed a new dural arteriovenous fistula in the posterior cranial fossa.

sinus. As reported by several other studies,<sup>9</sup> even without imaging the inferior petrosal sinus, the opening of the inferior petrosal sinus can usually be detected using a loach guidewire that is also inserted into the cavernous sinus. The lesson learnt from using femoral vein-IPSA was that a coaxial system should be used for operation, whereby a 120-cm 4-Fr catheter can be inserted into the 6-Fr guiding catheter and placed at the internal jugular vein bulb on the diseased side. The tip of the guiding catheter could then be pointed toward the anteromedial direction of the internal jugular vein bulb, and the loach guidewire can be used to explore the opening of the inferior petrosal sinus along the slope. Once the loach guidewire has found the opening, it can usually reach the cavernous sinus

through the inferior petrosal sinus, followed by the 120-cm 4F catheter entering into the inferior petrosal sinus. Force should strictly not be applied when entering into the inferior petrosal sinus; otherwise, the guiding catheter might stimulate, or be tangled with, the anterior and posterior abducens nerve of the Gurber ligament and cause damage. By using a blank roadmap, the loach guidewire was removed and the Echelon-14 microcatheter was introduced directly into the inferior petrosal sinus. The Echelon-14 microcatheter was passed through the gap where the cavernous sinus was located by coordinating with the Traxcess-14 microguidewire, and inserted into the dilated superior/inferior ophthalmic vein. The embolization treatment can then be performed after confirming the location of the microcatheter with super-selective angiography. All late patients treated through the venous approach at our center underwent surgery with this coaxial technique, which effectively solved the problem caused by the difficulty of super-selectively inserting the microcatheter into the inferior petrosal sinus through the jugular venous bulb, while also reducing the operation time and radiation dose. In addition, the 4F catheter provided relatively strong support to the microcatheter during embolization, which increased stability.

Spring coil embolization using the venous approach can achieve a certain level of targeting. The superior/inferior ophthalmic vein or the critical draining vein was the target point of our embolization. In general, the tip of the microcatheter was first placed at the opening of the superior/inferior ophthalmic vein or the critical draining vein, and spring coils of proper specifications according to the dilation level of vein can be used for dense embolization. This was done to avoid the inability of the fistula to be closed after the tip of the microcatheter was withdrawn into the cavernous sinus, resulting in non-improvement or even deterioration of postoperative symptoms. The fistula was the main target site of embolization. After closing the superior ophthalmic vein or the critical draining vein, the tip of the microcatheter was withdrawn into the gap where the fistula was located and was placed as close as possible to the fistula. The proper spring coils were chosen based on the size of the gap where the fistula was located, and dense embolization was performed at the fistula. During embolization, real-time angiography was performed to observe the fistula closing condition. Once the fistula was completely closed or the speed of the draining vein was obviously reduced and the therapeutic target had been achieved, it was also necessary to avoid excessive embolization, as the occurrence rate of excessive embolization,-related complications was reported to be as high as 10.7–39.4%.<sup>10,19</sup> In this study, one patient showed postoperative abducens nerve palsy that was considered to be related to excessive embolization, of the cavernous sinus.

In general, treatment of CSDAVFs with dural venous drainage involve careful planning, and healing can sometimes be very difficult to achieve when the disease is complex and there are multiple fistulas.<sup>2,4</sup> The six patients who underwent imaging of the draining vein after surgery represent such complex cases. For cases like these, combined embolization with coils and Onyx is likely to achieve better results.<sup>4</sup>

For the two patients with IPSA failure, one was switched to direct puncture of the superior ophthalmic vein, and healing embolization was successfully achieved once the microcatheter successfully reached the gap where the fistula was located, whereas the other patient was also successfully embolized using Onyx after switching to the arterial approach (Fig. 1). This indicates that when the fistula and draining vein are restricted in the anterior/anterior inferior gap, it can sometimes be difficult for the tip of the microcatheter to reach the gap where the lesion is located, thus causing difficulties in using IPSA to perform embolization. However, the fistula can mostly be healed if the microcatheter can reach the gap where the lesion is located through the venous approach. In such cases with a restricted fistula and draining vein, the blood-supplying artery is usually singular. If it is found that the microcatheter can easily approach the fistula through the arterial end after carefully analyzing the angioarchitecture, then the arterial approach can be used as the primary treatment approach.

The arterial approach of embolization for CSDAVF is a simple procedure with a low cost.<sup>6</sup> The use of a 5F guiding catheter is recommended when choosing embolization through the arterial approach in order to reach closer to the fistula, so that stronger support can be provided to the microcatheter when entering the location. In general, external carotid artery branches that are relatively thick, smooth, and provide easy approach to the fistula are used as the entrance for the microcatheter. The ascending pharyngeal artery and external carotid artery petrosus branch should be avoided, as these are blood-supplying vessels of multiple cranial nerves, which can be damaged if these two arteries are closed. Once it has been confirmed with super-selective angiography that the tip of the microcatheter is at or close to the fistula, the lesion features need to be reanalyzed and the anatomical structure needs to be re-examined, in order to anticipate the diffusion direction and reflux degree of Onyx. If the fistula has a large shunt flow and ectopic embolization of Onyx is a concern, partial filling of spring coils can be conducted, after which Onyx can be slowly injected for embolization with the guidance of the roadmap. Within the safe range anticipated, Onyx should be injected all at once so that it can be sufficiently diffused at the fistula. If the fistula can be diffused, the injection can be stopped once reflux is observed in the blood-supplying artery. If the fistula cannot be diffused, the injection should be stopped either when Onyx reflux occurs at the tip of the microcatheter or when Onyx spreads into the draining vein, and continued after a 30-s interval. Sufficient diffusion of Onyx at the fistula should be allowed through multiple injection-reflux-pause-reinjection cycles, to achieve as much blocking as possible. It is best to achieve satisfactory injection in one blood-supplying artery in order to reduce the number of operating procedures. Other blood-supplying arteries can be chosen for injection by using the same technique when necessary. The catheter should be withdrawn once the Onyx reflux exceeds 1.5-2 cm, and observation should normally be performed for no more than 2 min.<sup>15</sup>

Complications caused by Onyx injection through the arterial approach mainly occur because the blood-supplying vessels of cranial nerves are blocked and because of intracranial arterial embolization caused by dangerous anastomoses.<sup>16,17</sup> When the disease is complex, multiple fistulas as well as multiple plexiform vascular plexuses supplying blood exist, and the microcatheter cannot reach close to the fistula, it may be difficult for Onyx to diffuse the common draining venous pathway. As a result, healing may not be achieved and complications such as damaged cranial nerves due to blockage of blood-supplying vessels of the nerves can occur. Arterial embolization treatment is not suitable for such cases. One patient in this study showed facial palsy symptoms on the second day after surgery, which was suspected to be related to a blocked petrosus branch of the meningeal artery due to excessive Onyx reflux (the petrosus branch is the blood-supplying vessel of the facial nerves). The patient's facial palsy symptoms did not significantly improve throughout the 3-month follow-up after surgery. No event of intracranial artery embolization caused by dangerous anastomosis was observed in this study, which can be attributed to the comprehensive understanding of the anatomical structure of the lesion before injection, allowing effective control of Onyx diffusion.

Twenty-seven patients were followed up for 3–62 months, and the symptoms of 26 patients improved or disappeared. Only one 51-year-old female patient experienced tinnitus 3 months after surgery, which was diagnosed as anterior cranial fossa new-onset DAVF on DSA. Trauma, infection, or other possible causes were not found in our examination; however, this patient was experiencing menopause, indicating that estrogen level variations might be an initiating factor that caused DAVF, although the mechanism of DAVF onset still requires further investigation.

# Conclusion

On the basis of the angioarchitectural features of CSDAVF, the arterial approach can be selected as the primary treatment plan when the bloodsupplying artery and the fistula are relatively singular and when the microcatheter can easily reach close to the fistula through the artery. In contrast, the venous approach should be selected as the primary approach when the fistula is indistinguishable and blood is supplied by multiple arteries through small plexiform vessels. Choosing the optimal surgical approach may increase the success rate of intravascular CSDAVF surgeries and may help avoid complications.

# Disclosure

The authors report no conflicts of interest with respect to the materials and methods used in this study or the findings specified in this paper.

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