Case Report

Spontaneous Closure of a Cavernous Sinus Dural Arteriovenous Fistula with Spinal Perimedullary Drainage (Cognard V) during Attempted Transvenous Embolization

Abstract

The authors describe a patient with spontaneous closure of a spontaneous cavernous sinus dural arteriovenous fistula (CSDAVF), Cognard type V, during transvenous attempt. A 39-year-old woman experienced mild proptosis, redness of the left eye, and diplopia. Four months later, she developed left retro-orbital pain and left-sided headache. Cerebral angiography revealed the left CSDAVF exclusively draining into the superior petrosal sinus with subsequent drainage into the veins surrounding the medulla oblongata, and finally into the perimedullary spinal veins, classified as Cognard type V. The feeders arise from the dural branches of both the left external and internal carotid arteries. Following 2 h period of attempted transvenous embolization, the fistula disappeared spontaneously. Follow-up angiography obtained 6 months later confirmed complete resolution of the CSDAVF. At 2-year follow-up, the patient has remained clinically asymptomatic. The mechanism of thrombosis in this fistula related to the endovascular procedure. We speculated that putting the wire tip in the draining vein may induce the spontaneous thrombosis in the venous side. In addition, precipitating factors may include small, low-flow fistula, and pre-existing thrombosis.

Keywords: Cavernous sinus, Cognard type V, dural arteriovenous fistula, spontaneous closure, thrombosis, transvenous embolization

Introduction

Intracranial dural arteriovenous fistulas (DAVFs) are abnormal arteriovenous connections located within the dura matter, commonly near the venous sinuses. The arterial supply usually arises from dural or meningeal arteries. Dural branches from the pial arteries are rare. Venous drainage may occur into the dural sinuses, meningeal veins, osteodural veins, and/or retrogradely leptomeningeal veins. These fistulas are an acquired disease.^[1] Symptoms of the patients usually relate to location of the fistulas and venous drainage patterns.^[2,3]

The cavernous sinus (CS) is the second-most common location of intracranial DAVFs.^[4,5] The etiology of most CSDAVFs, abnormal communications between dural arteries and the CS, remains unclear. As explained by Newton and Hoyt,^[6] the occurrence of the fistulas may be caused by rupture of the thin-walled dural arteries traversing the CS, predisposed by vascular disease, straining, or trauma. Spontaneous CSDAVFs, low

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Based on the pattern of venous drainage by Cognard *et al.*,^[4] intracranial DAVFs were classified into five types, including type I: fistulas drain into the main sinus with antegrade flow; type IIa: fistulas drain

How to cite this article: lampreechakul P, Wangtanaphat K, Lertbutsayanukul P, Wattanasen Y, Siriwimonmas S. Spontaneous closure of a cavernous sinus dural arteriovenous fistula with spinal perimedullary drainage (Cognard V) during attempted transvenous embolization. Asian J Neurosurg 2019;14:1268-74. Prasert lampreechakul, Korrapakc Wangtanaphat, Punjama Lertbutsayanukul¹, Yodkhwan Wattanasen¹, Somkiet Siriwimonmas²

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retrograde flow into the sinus(es) only; type IIb: fistulas drain retrograde flow to cortical vein(s) only; type IIa + b: fistulas drain retrograde flow into sinus(es) and cortical vein(s); type III: fistulas drain direct cortical venous drainage; type IV: fistulas drain direct cortical venous drainage with venous ectasia; and type V: fistulas drain into the spinal perimedullary veins.

Spontaneous closure of intracranial DAVFs is rare and usually occurs in benign fistulas, i.e., Cognard type I and IIa. The occurrence of spontaneous regression in cranial DAVFs with cortical venous reflux or higher type of Cognard classification is extremely rare.^[11,12]

The authors reported a patient with spontaneous disappearance of CSDAVF predominately draining into spinal perimedullary veins, i.e., Cognard V, during attempted transvenous embolization.

Case Report

A 39-year-old woman with a medical history of migraine for 1 year, noticed mild proptosis, redness of the left eye, and diplopia. There was no history of trauma. She went to the local hospital and was diagnosed of conjunctivitis, treated by medical treatment without improvement. Four months later, she developed left retro-orbital pain and left-sided headache. The patient was sent to another local hospital and obtained magnetic resonance imaging (MRI). The study showed abnormal bulging of the left CS with dilated left superior ophthalmic vein (SOV) [Figure 1]. Provisional diagnosis was the left CSDAVF. She was transferred to Prasat Neurological Institute for further investigation and treatment. On neurological examination, there was only mild hyperreflexia of both extremities without muscle weakness. Ophthalmic examination revealed mild proptosis, dilated episcleral veins, and a slight abducens nerve paresis of the left eye. Visual acuity was 20/20 in both eyes. Fundoscopic examination showed venous stasis retinopathy in the left eye. IOP was 8.6 mmHg in

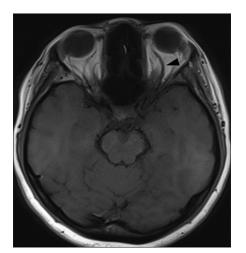


Figure 1: Axial T1-weighted magnetic resonance imaging of the brain shows an enlarged left superior ophthalmic vein (arrowhead)

the right eye and 13.2 mmHg in the left eye. Left internal carotid angiography with angiographic computerized tomography in maximum intensity projection-reformatted images revealed a DAVF shunting into the posterior CS, draining exclusively into the enlarged left superior petrosal sinus (SPS), left petrosal vein, left lateral medullary vein, enlarged anterior spinal vein, contralateral lateral medullary vein, right petrosal vein, right SPS, and finally into the right transverse sinus and inferior petrosal sinus (IPS). There was no opacification of the posterior segment of the left SPS. The reflux of venous outflow into the anterior portion of the left CS and SOV was seen on the late venous phase, probably representing partially thrombosed left CS and SOV [Figures 2-4]. Also, late arterial phase of the left ICA injection disclosed the cervical perimedullary veins draining into the epidural veins at the level of C1 and C3. There was also the venous drainage into cerebellar veins [Figure 5]. In addition, the fistula received blood supply from the right ICA, i.e., the meningohypophyseal trunk, and the left ECA, i.e., the artery of foramen rotundum and middle meningeal branches [Figure 6]. Due to a large volume of an interventional neuroradiology service at our tertiary referral center, endovascular treatment was scheduled in the next 3 months. Cerebral angiography obtained 3 months later disclosed the existence of the fistula which remained unchanged. Under general anesthesia, the first transvenous embolization via the left IPS without heparinization was unsuccessful because of the inability to deliver the microcatheter into the left CS. The patient refused further surgical treatment. Four months later, another transvenous embolization via the same route was performed again under general anesthesia. Furthermore, it was difficult to navigate the microcatheter into the left CS. We could deliver the microguidewire into the left petrosal vein coming from the IPS, but the microcatheter could not follow the microguidewire to the CS [Figure 7a]. Cerebral angiography, obtained 2 h after attempting embolization, revealed the absence of early opacification of the venous drainage of the left CS with only prominent dural arterial supply from the cavernous segment of the left internal carotid, middle meningeal, and internal maxillary arteries [Figure 7b and c]. There was no hypotension during the procedure.

On 6-month follow-up after spontaneous disappearance of the left CSDAVF, the patient no longer had headache. Proptosis and cranial nerve dysfunction had completely resolved. Follow-up IOP was 7.6 mmHg in the right eye and 8.9 mmHg in the left eye. Fundoscopic examination revealed complete resolution of venous stasis retinopathy. Cerebral angiography also confirmed complete resolution of the fistula [Figure 8]. At 2-year follow-up, the patient has remained clinically asymptomatic.

Discussion

Spontaneous CSDAVFs often have been initially misdiagnosed, especially caused by the absence of a

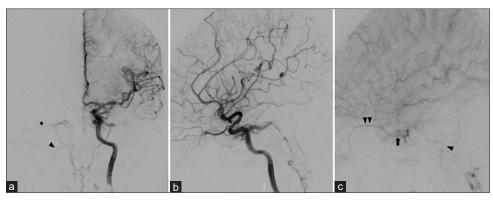


Figure 2: Cerebral angiography obtained 4 months after initial symptoms. Anteroposterior (a) and lateral in the arterial (b) and late venous (c) phase views of the left internal carotid artery injection reveal a left-sided dural arteriovenous fistula shunting into the posterior cavernous sinus, draining exclusively into an enlarged left superior petrosal sinus, left petrosal vein, left lateral medullary vein, enlarged anterior spinal vein, contralateral lateral medullary vein, right petrosal vein, right superior petrosal sinus (asterisk), and finally into the right transverse sinus and inferior petrosal sinus (arrowhead). There is no opacification of the posterior segment of the left superior petrosal sinus. The reflux of venous outflow into the posterior portion of the left cavernous sinus (arrowheads) is seen on the late venous phase, probably representing partially thrombosed left cavernous sinus and superior ophthalmic vein

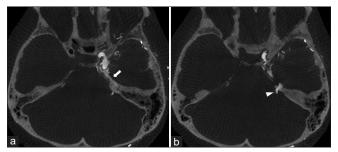


Figure 3: Axial maximum intensity projection-reformatted sequential images of angiographic computerized tomography of the brain (a and b) clearly show venous drainage of the left cavernous sinus dural arteriovenous fistula into an enlarged left superior petrosal sinus (arrow) along the superior angle of the petrous part of the left temporal bone, directly draining into a left petrosal vein (arrowhead)

bruit.^[13] In addition, our case presented with mild proptosis, redness of the left eye, and diplopia without bruit, leading to misdiagnosis from the previous hospital.

The findings of incomplete opacification of the CS and abnormal retrograde venous drainage are highly suspicious of partial thrombosis of the CS.^[9] The angiography of our case showed no filling of the SOV in early arterial phase incompatible with her symptoms, probably representing preexisting thrombosis. Headache may be the sign of partial thrombosis of the CS.^[10] It is possible that her subsequently developing headache may be the sign of CS thrombosis.

The CSDAVFs can cause venous congestion of the brainstem, leading to catastrophic consequences.^[14-17] Hypoplasia or occlusion of posterior segment of the SPS, divided into anterior and posterior segments, may accelerate the venous reflux into the petrosal vein and subsequent drainage into the veins of brainstem and posterior fossa.^[14,18,19] Retrograde venous drainage into the brainstem and cerebellum from the CSDAVFs may result in venous infarction and/or intracranial hemorrhage.^[20]

The CSDAVF in our case was supplied by dural branches of both the ICA and ECA with predominantly drainage into the perimedullary veins via SPS and petrosal vein, corresponding with a classification of a Cognard type V DAVF.

Cognard type V accounts for 5.9% of all intracranial DAVFs and may experience myelopathy, bulbar palsy, and respiratory dysfunction, if left untreated.[4,21] Regarding the study of relation between clinical presentation and angiographic findings of intracranial DAVFs with spinal venous drainage by Brunereau et al.,[22] the authors found that patients without myelopathy had the venous drainage limited to the cervical region, whereas the venous drainage in patients with myelopathy descended toward the conus medullaris. Due to the perimedullary veins draining into the epidural veins through a medullary-radicular vein, the occurrence of spinal cord congestion or hypertension was prevented. Similarly, our case had no myelopathic symptoms probably due to the drainage of the perimedullary veins into the epidural veins and the contralateral transverse and IPS. We previously reviewed and elucidated the venous drainage of the spine and spinal cord in other interesting case reports.^[23,24]

Spontaneous CSDAVFs have a relatively high incidence of spontaneous regression.^[10,13,25-27] Then, management of these type of fistulas should be conservative unless the patients have progressive loss of vision. Thrombosis of venous outlet of a DAVF may either reroute drainage into cortical veins, leading to hemorrhagic complications, or induce spontaneous regression of DAVFs. However, it is difficult to predict which DAVF will regress or transform into a more dangerous type.^[2] van Dijk *et al.*^[11] reported two patients harboring CSDAVFs with long-term persistent cortical venous reflux, i.e., Cognard type IIa + b. Without treatment, spontaneous cure occurred in one patient after 14.5-year follow-up. Another patient had moderate disability 9.1 years later.

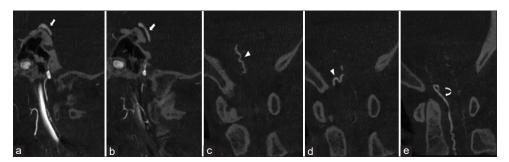


Figure 4: Sagittal maximum intensity projection-reformatted sequential images, from the left side to the midline, of angiographic computerized tomography of the craniocervical junction (a-e) demonstrate an enlarged left petrosal vein (arrow) caudally draining into the lateral medullary vein (arrowhead), and finally joining with a dilated anterior median spinal vein (curve arrow)

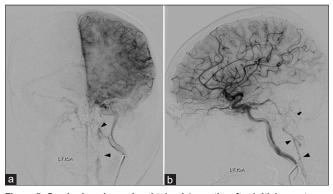


Figure 5: Cerebral angiography obtained 4 months after initial symptoms. Anteroposterior (a) and lateral (b) views of the left internal carotid angiography, late arterial phase, reveal the perimedullary veins draining into the epidural veins (arrowheads) at the level of C1 and C3. There is also the venous drainage into cerebellar veins (arrow)

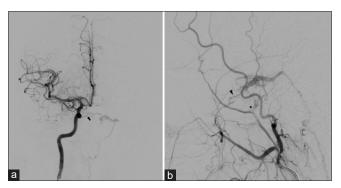


Figure 6: Cerebral angiography obtained 4 months after initial symptoms. (a) Anteroposterior view of the right internal carotid artery injection shows a left-sided dural carotid-cavernous fistula supplied by medial clival branch (arrow) of the right meningohypophyseal trunk of the cavernous segment of the right internal carotid artery via midline anastomotic branches. (b) Lateral view of the left external carotid artery injection reveals additional blood supply from the artery of foramen rotundum (arrowhead) and middle meningeal branches (asterisk)

The goal of treatment of Cognard type V DAVFs must be complete occlusion.^[4] Incomplete treatment allows recruitment of collateral vessels and persistent risk of aggressive symptoms.^[3] Without approaches for transarterial or transvenous embolization, direct embolization with embolic materials through open surgery may be another option.^[17] The most effective treatment option of CSDAVFs is primarily endovascular treatment

by transarterial or transvenous approach.^[28] Due to the risk of embolic complications associated with transarterial embolization of CSDAVFs, transvenous approach through the IPS using coils and/or liquid embolic materials is the preferred method.^[3,8,9,27,29] Even though IPS is not opacified by angiography, transvenous catheterization into the CS can be performed successfully through occluded venous segment.^[8,29,30]

Due to a high incidence of spontaneous obliteration, patients with CSDAVFs showing signs of venous thrombosis at angiography should be treated by conservative management.^[7] In our case, however, subsequent diversion of venous drainage into perimedullary veins may be aggravated following complete occlusion or thrombosis of the SOV. Thus, we decided to treat our patient due to a possibility of developing unpredictable aggressive neurological symptoms. Despite the absence of ipsilateral IPS drainage, we preferred to use transvenous embolization with coiling because we have more experience on this technique and considered surgery as the last option. Fortunately, the fistula disappeared spontaneously during difficult endovascular procedure.

Endovascular procedure-related spontaneous closure of intracranial AVFs has been scarcely reported. In the large series of Lewis et al.[31] the authors reported spontaneous closure of three direct CCFs after unsuccessful attempts to place a detachable balloon. Similarly, closure of the fistula during attempted embolization without placement of a balloon was reported in previous studies.^[32,33] In addition, Uchino et al.[34] reported spontaneous obliteration of small traumatic direct CCF following unsuccessful transarterial balloon embolization. They demonstrated that the shunt flow reduced significantly during the procedure and speculated that repeated transient reduction and stagnancy of the blood flow may promote secondary thrombosis in this patient. This concept was demonstrated by the report of Whang et al.[35] presenting incidentally formed thrombus during difficult navigation of microcatheter and microguidewire through the small-sized fistula, which induced sudden closure of the fistula. In addition, Berenstein and Lasjaunias^[36] showed spontaneous closure of the vertebro-vertebral fistula occurred while attempting

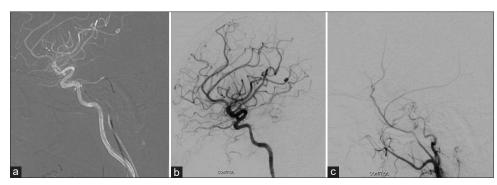


Figure 7: (a) During superselective transvenous catheterization of the left cavernous sinus. Lateral view of the left internal carotid artery injection during angiographic road-mapping demonstrates the microguidewire navigated into the left petrosal vein through the left superior petrosal sinus and the microcatheter remaining in a left inferior petrosal sinus. Lateral views of the left internal (b) and external (c) carotid artery angiography, obtained 2 h after attempting embolization, reveal the absence of early opacification of the venous drainage of the left cavernous sinus with only prominent dural arterial supply from the cavernous segment of the left internal carotid, middle meningeal, and internal maxillary arteries

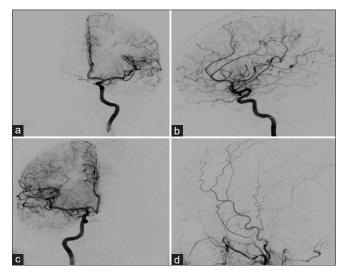


Figure 8: Cerebral angiography obtained 6 months after the second attempt of transvenous embolization. Anteroposterior (a) and lateral (b) views of the left internal carotid artery, anteroposterior view of the right internal carotid artery (c), and lateral view of the left external carotid artery (d) injections confirm complete resolution of the left cavernous sinus dural arteriovenous fistula

balloon embolization. Direct CCFs closed spontaneously not only after a failed transarterial approach, but also following an attempted transvenous approach.^[37]

There are several possible mechanisms, in which the DAVFs close spontaneously, proposed in previous studies. Luciani *et al.*^[38] reviewed the literature of spontaneous closure of DAVFs and divided into two types, i.e., posttraumatic and spontaneous. Most of the posttraumatic DAVFs were Cognard type I small fistulas, probably more likely to occlude spontaneously. In addition, posttraumatic vessels tend to develop scar tissue during the healing process, while spontaneous group has two mechanisms of spontaneous closure, including sinus thrombosis and intracranial hemorrhage. All Cognard type III DAVFs were diagnosed following intracranial hemorrhage. Regarding literature review from published data of spontaneous closure of intracranial DAVFs by Al-Afif *et al.*,^[12] this situation can occur in the fistulas classified

as Cognard type I, IIa, IIa + b, IIb, III, and IV. The most common spontaneously resolving lesions were type I. Warren et al.^[39] reported another rare precipitating factor, i.e., spontaneous resolution of Cognard type IIa DAVF following spontaneous recanalization of the occluded sinus. Voormolen et al.^[40] reported spontaneous closure of Cognard type IV intracranial DAVFs, draining directly into the cortical venous drainage with several ectasias in its course, in two middle-aged women. They speculated that the predisposing factors were small fistula, single-dilated cortical vein, and the specific contrast agent used during diagnostic angiography. Spontaneous resolution of Cognard type IIb and III intracranial DAVFs, with cortical venous drainage, may be caused by hemorrhage and the mass effect related to hematoma.[41] Vasospasm may also be a potential mechanism related to bleeding. To the best of our knowledge, there was no previous case report of spontaneous closure of Cognard type V intracranial DAVF.

Spontaneous obliteration of low-flow CSDAVFs may result from partial or complete thrombosis of the CS and/or its tributaries.^[7] Patients harboring CSDAVFs without cortical venous drainage may achieve complete obliteration by external manual carotid compression. Simultaneous compression of the carotid artery and jugular vein may temporarily equalize the pressure between the carotid inflow to the fistula and the CS outflow, leading to stasis and thrombosis.^[42] CSDAVFs may resolve shortly following diagnostic cerebral angiography.^[43] The injection of contrast media may exaggerate the adhesion to and invasion of the venous wall by leukocytes and possibly contribute to thrombus formation.[44] Furthermore, cervical carotid angiography can produce complications, including arterial spasm or dissection. In addition, some patients underwent angiography under general anesthesia, leading to lowering systemic blood pressure, resulting in reduction of the flow in the fistula.^[45] Spontaneous cure of the CSDAVFs may relate to pregnancy or delivery due to changes in blood coagulation and direct effect of estrogens on the vascular wall.^[46] Single venous drainage is another predisposing factor of complete remission.^[10]

In the present case, spontaneous disappearance of the fistula seems to correlate with endovascular procedure, i.e., putting the wire tip in the draining may induce the spontaneous thrombosis in the venous side. In addition, precipitating factors may include small, low-flow fistula, and preexisting thrombosis. However, contrast agents may not play an important role in the spontaneous closure of the fistula in our case due to the persistence of the fistula 7 months following the first angiography. To confirm the durability of complete cure of the fistula, long-term follow-up is necessary.

El Asri *et al.*^[47] conducted a systemic review of literature on patients with intracranial DAVFs with perimedullary drainage; they found that the poor outcomes were correlated to the presence of abnormal hyperintensity of the brainstem on T2-weighted imaging. Fortunately, there was no this finding on MRI in our case.

Conclusion

The authors reported endovascular procedure-related spontaneous closure of a Cognard type V CSDAVF in a young adult woman. During transvenous attempt via the IPS, putting the wire tip in the draining vein can even induce the spontaneous thrombosis in the venous side. In addition, precipitating factors may include small, low-flow fistula, and preexisting thrombosis.

Consent

The patient has given consent to be enrolled and has her data published.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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