



Case Report

Spontaneous complete regression of malignant cavernous sinus dural arteriovenous fistula following partial transarterial embolization with liquid embolic material: Report of two cases

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ABSTRACT

Background: Spontaneous complete regression of malignant cavernous sinus dural arteriovenous fistulas (CSDAVFs) following partial transarterial embolization is an extremely uncommon phenomenon. The mechanism responsible for this condition remains unclear.

Case Description: The authors describe two cases of malignant CSDAVFs (Cognard IIB and V) treated by partial transarterial embolization with liquid embolic agents after unsuccessful transvenous embolization through various routes. Follow-up cerebral angiography in these cases confirmed complete resolution of the fistulas.

Conclusion: In our two patients harboring low-flow CSDAVFs with preexisting thrombosis of the cavernous sinus (CS), it is possible that some portions of the liquid embolic materials could migrate into the fistulas, inducing thrombosis within the CS.

Keywords: Cavernous sinus dural arteriovenous fistula, Intracranial dural arteriovenous fistula, Spontaneous closure, Spontaneous regression, Transarterial embolization

INTRODUCTION

Cavernous sinus dural arteriovenous fistulas (CSDAVFs) are abnormal communications between dural arteries and the cavernous sinus (CS). Spontaneous CSDAVFs, low flow, and low pressure, commonly appear in middle-aged females, and symptoms of the patients usually relate to venous drainage patterns.^[18]

Based on the pattern of venous drainage by Cognard *et al.*,^[5] intracranial DAVFs were classified into five types, including type I: fistulas drain into the main sinus with antegrade flow; type IIa: fistulas drain 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

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drainage with venous ectasia; and type V: fistulas drain into the spinal perimedullary veins.

At present, a transfemoral transvenous approach through the ipsilateral inferior petrosal sinus (IPS) is recommended as the first-line treatment of CSDAVFs.^[11] Transarterial embolization of CSDAVFs is usually difficult and infrequently used due to small feeding branches and the risk of ischemic stroke and cranial nerve palsy.^[7,16,17] If an unsuccessful transvenous approach, transarterial embolization may be considered as an alternative approach in selected patients.^[14] Spontaneous regression of CSDAVFs following partial transarterial embolization is rare and usually occurs in benign fistulas, that is, Cognard type I and IIa.^[14,21] The authors reported two patients with spontaneous obliteration of malignant DAVFs (Cognard IIb and V) after partial transarterial embolization of these fistulas with liquid embolic agents.

CASE DESCRIPTION

Case 1

A 32-year-old woman noticed headache with proptosis and redness of the right eye for 1 month. There was no history

of trauma. She went to the local hospital and received contrast-enhanced cranial computed tomography (CT) scan that showed abnormal bulging of the right CS with dilated right superior ophthalmic vein (SOV). The provisional diagnosis was the right CSDAVF. She was transferred to our institute for further investigation and treatment. Cerebral angiography revealed the right CSDAVF supplied from the right artery of the foramen rotundum, accessory meningeal artery, middle meningeal artery (MMA), and dural branches of the bilateral cavernous segment of internal carotid arteries (ICAs) with drainage into the right mildly dilated SOV and superior petrosal sinus (SPS) with subsequently draining into the right sigmoid sinus. In addition, drainage was through the right lateral medullary veins into the dilated spinal perimedullary veins, corresponding with a classification of a Cognard type V DAVF [Figure 1]. Under general anesthesia, attempts for transvenous embolization of the fistula through the right SPS and occluded IPS were unsuccessful. Therefore, transarterial embolization with N-butyl cyanoacrylate (NBCA) through the right accessory and meningeal arteries was performed with the result of incomplete obliteration of the fistula [Figure 2]. Another session of embolization was scheduled for the next month. However, she denied further treatment because her symptoms disappeared 2 weeks after

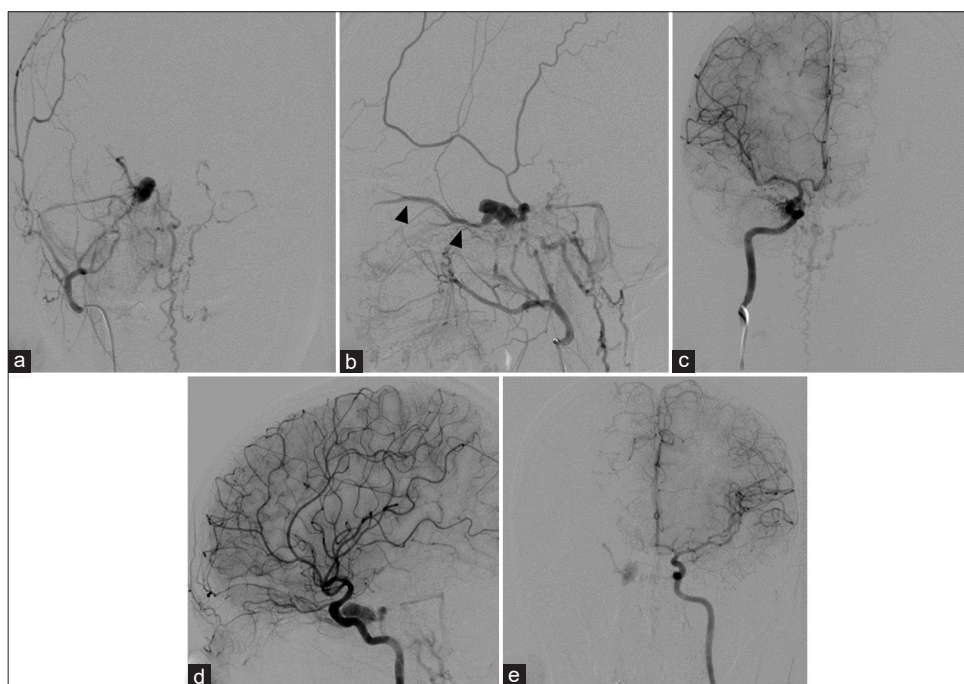


Figure 1: (a) Anteroposterior (AP) and (b) lateral views of the right external carotid artery injections show the right cavernous sinus dural arteriovenous fistula supplied from the right artery of the foramen rotundum, accessory meningeal artery, middle meningeal artery with drainage into the right superior ophthalmic vein (SOV), superior petrosal sinus and spinal medullary veins. There is partial filling of the right SOV (arrowheads) incompatible with the patient's ocular symptoms, probably representing pre-existing thrombosis. (c) AP and (d) lateral views of the right internal carotid artery (ICA) injections reveal the vascular supply from meningohypophyseal trunk (MHT). (e) AP view of the left ICA injection demonstrates the clival branch from the left MHT supplying the fistula.

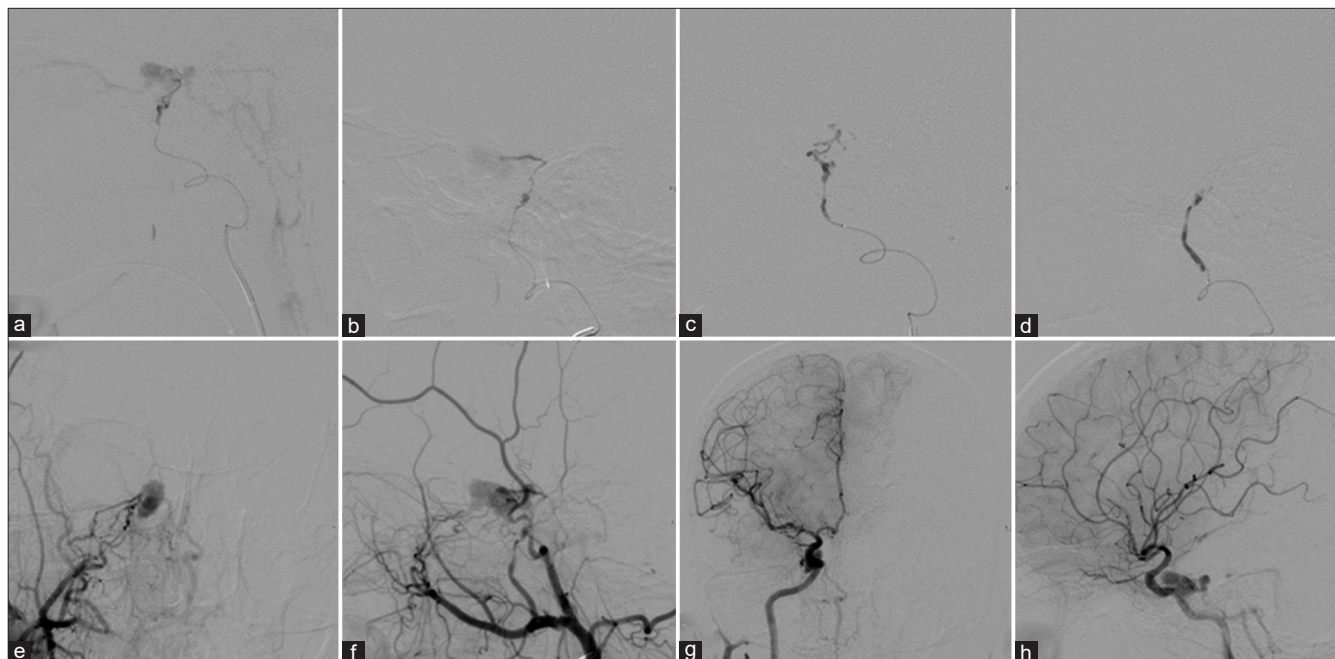


Figure 2: Lateral views of the right (a) accessory meningeal artery and (b) middle meningeal artery injections before embolization. During glue embolization from the right (c) accessory meningeal artery and (d) middle meningeal artery. (e) Anteroposterior (AP) and (f) lateral views of the right external carotid artery and (g) AP and (h) lateral views of the right internal carotid artery injections demonstrate incomplete obliteration of the fistula.

embolization. Six months following endovascular treatment, cerebral angiography was obtained and demonstrated the disappearance of the fistula [Figure 3]. At the 3-year follow-up, she remained clinically asymptomatic.

Case 2

A 52-year-old woman went to the local hospital due to proptosis and redness of the right eye for 4 days. One month earlier, she noticed tinnitus in her right ear. There was no history of trauma. Contrast-enhanced CT scan of the brain revealed abnormal bulging of the right CS with dilated right SOV. The patient was diagnosed with the right CSDAVF and sent to our institute. Unfortunately, she could not come to our institute at that moment due to the COVID-19 pandemic. Two months later, she developed a mild headache and progression of her eye symptoms for 1 week. Later, her ocular symptoms and tinnitus disappeared totally. Six months later, the patient came to our institute for further investigation and treatment. Cerebral angiography showed the right CSDAVF fed by the right artery of the foramen rotundum, accessory meningeal artery, MMA, and dural branches of the bilateral cavernous segment of ICAs with drainage into the right dilated superficial middle cerebral and Labbé veins, corresponding with a classification of a Cognard type IIb DAVF [Figure 4]. To prevent intracerebral hemorrhage from the cortical venous reflux, she was advised to treat this fistula. Under general anesthesia, attempts for

transvenous embolization of the fistula through the right occluded IPS SPS and the vein of Labbé were unsuccessful. Therefore, transarterial embolization with Onyx through the right accessory meningeal artery was performed with some part of Onyx filling into the CS. Control angiography after embolization demonstrated incomplete obliteration of the fistula [Figure 5]. Another session of embolization was scheduled, and cerebral angiography obtained 4 months later revealed complete regression of the fistula [Figure 6]. At the 2-year follow-up, the patient remained clinically asymptomatic.

DISCUSSION

Due to the risk of embolic complications associated with transarterial embolization of CSDAVFs, a transfemoral transvenous approach through the ipsilateral IPS is recommended as the first-line access, whether the IPS is patent or occluded.^[10] Following an unsuccessful approach through ipsilateral IPS, the cavernous sinus may be accessed through various routes, including contralateral IPS, SPS, facial vein, pterygoid plexus, or superficial middle cerebral vein.^[11] Transorbital access by direct puncture or surgical exposure of the distal SOV is another viable option. However, this technique is the most invasive modality of embolization and carries a greater risk of procedural complications, such as orbital hemorrhage.^[17] If unsuccessful transvenous approach, transarterial embolization may be considered as an

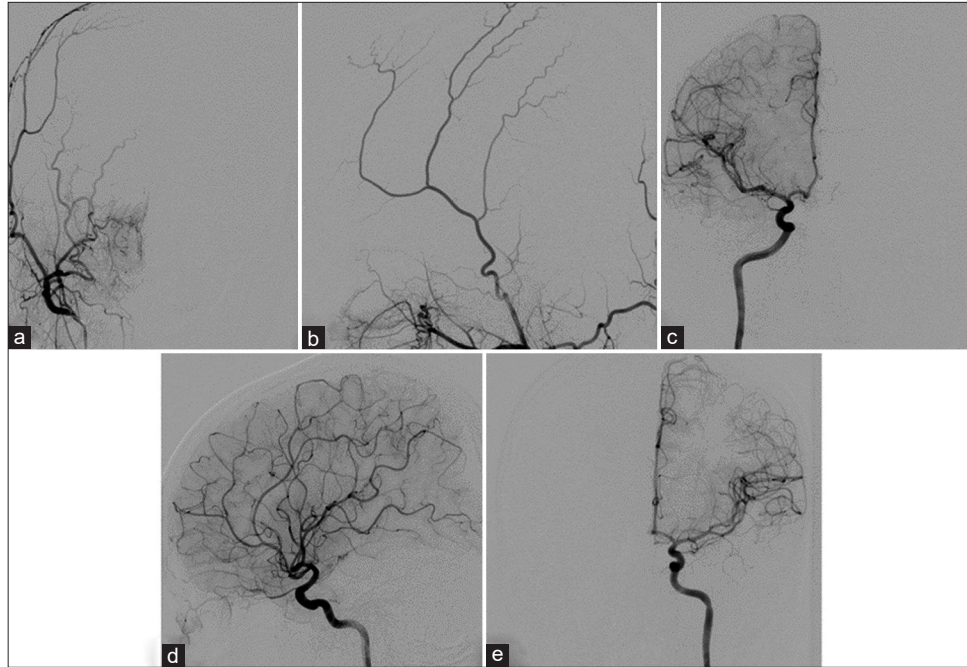


Figure 3: Cerebral angiography obtained 6 months after embolization. (a) Anteroposterior (AP) and (b) lateral views of the right external carotid artery, (c) AP and (d) lateral views of the right internal carotid artery (ICA), and (e) AP view of the left ICA injections confirms complete obliteration of the right cavernous sinus dural arteriovenous fistula.

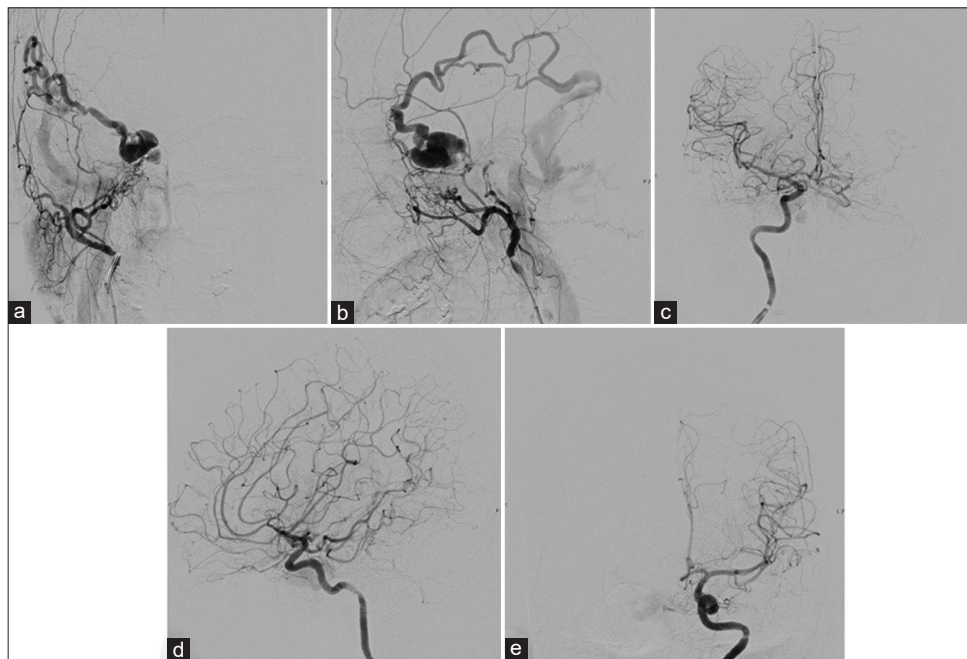


Figure 4: (a) Anteroposterior (AP) and (b) lateral views of the right external carotid artery and (c) AP and (d) lateral views of the right internal carotid artery (ICA) injections show the right cavernous sinus dural arteriovenous fistula supplied from the right artery of the foramen rotundum, accessory meningeal artery, middle meningeal artery, and meningohypophyseal trunk (MHT) with drainage into the right dilated superficial middle cerebral and Labbé veins. There is no drainage into the right superior ophthalmic vein incompatible with the patient's ocular symptoms, probably representing pre-existing thrombosis. (e) AP view of the left ICA injection demonstrates the clival branch from the left MHT supplying the fistula.

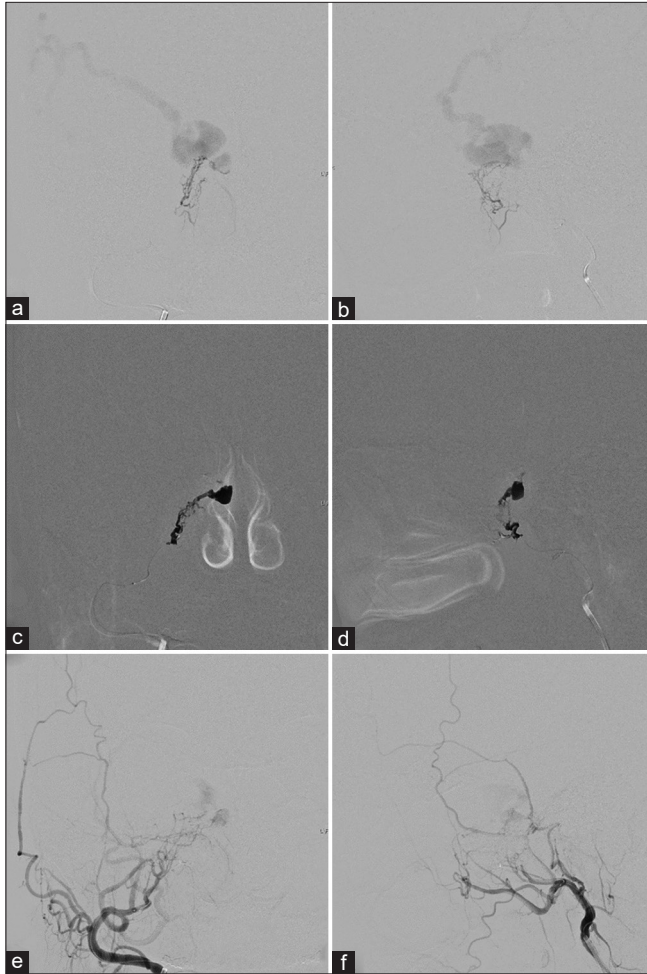


Figure 5: (a) Anteroposterior (AP) and (b) lateral views of the right accessory meningeal artery injection show the right cavernous sinus dural arteriovenous fistula with drainage into the right dilated superficial middle cerebral and Labbé veins. (c) AP and (d) lateral views during Onyx injection. (e) AP and (f) lateral views of the right external carotid artery injection reveal incomplete obliteration of the fistula.

alternative approach.^[19] Due to dangerous anastomosis of the external-internal carotid artery and potential vascular supply to the vasa nervorum of the lower cranial nerves, transarterial embolization of CSDAVFs should be the last resort in selected patients. In addition, superselective angiography with a microcatheter should be evaluated for the presence of potential anastomosis. Liquid embolic material can be used either with NBCA or Onyx through dural branches of the external carotid artery including MMA, accessory meningeal artery, or ascending pharyngeal artery.^[7,14] Protective balloon inflation at the cavernous segment of ICA may be useful for avoiding dangerous anastomosis embolization.^[16] In our cases, transvenous access to the fistulas through IPS and possible routes were attempted but failed. Without other accessible routes, we decided to use transarterial embolization

with liquid embolic materials, but it was unsuccessful to achieve complete obliteration of fistulas.

Over the years, polyvinyl alcohol (PVA) particles and NBCA have frequently been used as embolic agents for CSDAVFs.^[8,14,23] At present, ethylene-vinyl alcohol (EVOH) copolymer-based liquid embolic agent, such as Onyx, has widely been used for intracranial DAVFs or hypervascular lesions due to slow polymerization and nonadhesive properties providing reduced risk of microcatheter entrapment than NBCA and prolonged embolization time during the embolization resulting in the more controlled injection of the agent.^[6,12,14] We preferred EVOH as the first choice of liquid embolic material for CSDAVFs. In our first case, we used NBCA due to no EVOH available at that time.

CSDAVFs without cortical venous drainage have a relatively high incidence of spontaneous regression.^[4,13,15] However, the occurrence of spontaneous closure in cranial DAVFs with cortical venous reflux or a higher type of Cognard classification is extremely rare.^[3,10] Fortunately, spontaneous complete regression of the fistulas, including Cognard type IIb and V, occurred in our 2 cases.

To the best of our knowledge, spontaneous obliteration of malignant CSDAVFs after partial embolization through a transarterial route with liquid embolic materials has never been described in the available and relevant literature. In general, uncured CSDAVFs after transarterial embolization required further other therapeutic modalities, including transvenous and surgical approaches.^[1,2,11]

The mechanism of further spontaneous regression of malignant CSDAVFs following partial transarterial embolization with liquid embolic material remains unclear.^[4,9] After transarterial embolization, the abrupt changes in arterial flow may influence the local circulation and alter the venous drainage pattern, probably resulting in a more aggressive natural history. The recurrence of the fistulas may occur due to recruiting of new feeding arteries.^[21]

Spontaneous obliteration of low-flow CSDAVFs may result from partial or complete thrombosis of the CS and/or its tributaries.^[22] CSDAVFs may resolve shortly following diagnostic cerebral angiography.^[24] The injection of contrast media may exaggerate the adhesion to and invasion of the venous wall by leukocytes and possibly contribute to thrombus formation.^[20] In addition, cervical carotid angiography can produce complications, including arterial spasm or dissection. Furthermore, performing cerebral angiography under general anesthesia may lead to lowering systemic blood pressure, resulting in the reduction of the flow in the fistula.^[9] Interestingly, Iampreechakul *et al.*^[10] reported spontaneous closure of CSDAVF with spinal perimedullary drainage (Cognard V) during attempted transvenous embolization. They speculated that putting the wire tip into

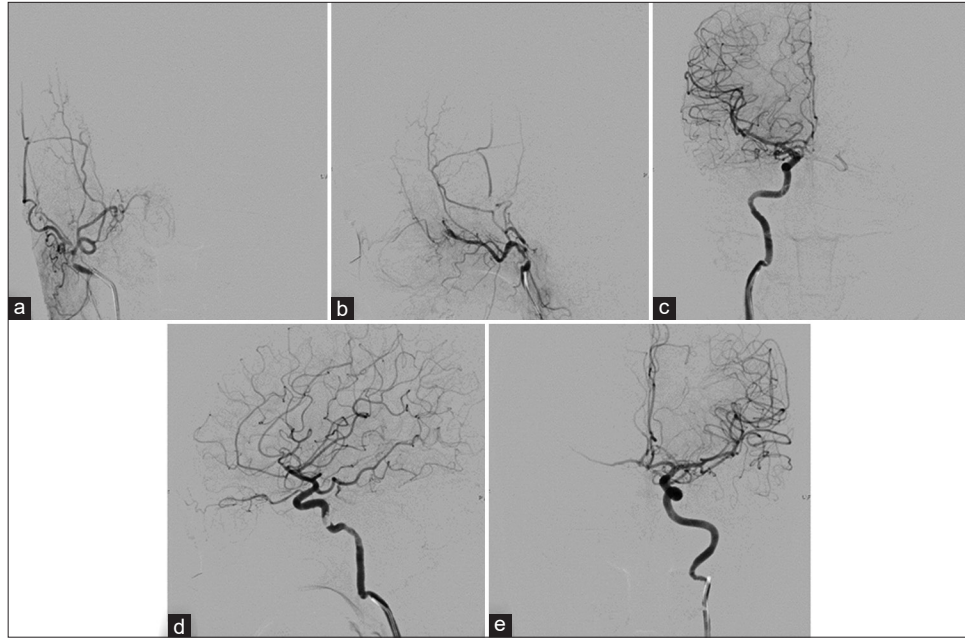


Figure 6: Cerebral angiography obtained 6 months after embolization. (a) Anteroposterior (AP) and (b) lateral views of the right external carotid artery, (c) AP and (d) lateral views of the right internal carotid artery (ICA), and (e) AP view of the left ICA injections reveal complete regression of the right cavernous sinus dural arteriovenous fistula.

the draining vein may induce the spontaneous thrombosis on the venous side.

The findings of incomplete opacification of the CS and abnormal retrograde venous drainage are highly suspicious of partial thrombosis of the CS.^[23] The angiography of our cases showed partial filling of the SOV in the artery phase incompatible with their symptoms and findings on the CT scan, probably representing pre-existing thrombosis. Headache may be a sign of partial thrombosis of the CS.^[15] It is possible that their subsequently developing headache may be a sign of CS thrombosis. In addition, following partial transarterial embolization with liquid embolic materials in our cases, we speculated that some portions of liquid embolic agents could migrate into the fistulas, inducing further thrombosis within the CS, resulting in complete closure of the fistulas.

Importantly, long-term follow-up of the incompletely cured patient is required for validation of the durability of the complete cure of the fistula.^[21] Our two cases have remained clinically asymptomatic during at least 2 years of follow-up.

CONCLUSION

Spontaneous regression of malignant CSDAVFs following partial transarterial embolization is an exceedingly rare condition. In our two patients harboring low-flow CSDAVFs with preexisting thrombosis of the CS, it is possible that some parts of the liquid embolic materials could penetrate

the fistulas, inducing thrombosis within the CS, resulting in complete closure of the fistulas.

Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of Artificial Intelligence (AI)-Assisted Technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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