



Case Report

A case of recurrent cavernous sinus dural arteriovenous fistula arising after superselective shunt occlusion and detected by venous arterial spin labeling

Hiromu Sunada¹, Ryosuke Maeoka^{1,2}, Ichiro Nakagawa², Hiroyuki Nakase², Hideyuki Ohnishi¹

¹Department of Neurosurgery, Ohnishi Neurological Center, Akashi, Hyogo Prefecture, ²Department of Neurosurgery, Nara Medical University, Kashihara, Nara Prefecture, Japan.

E-mail: Hiromu Sunada - h.sunada@onc.akashi.hyogo.jp; *Ryosuke Maeoka - r.maeoka@narmed-u.ac.jp; Ichiro Nakagawa - nakagawa@nmu-gw.narmed-u.ac.jp; Hiroyuki Nakase - nakasehi@narmed-u.ac.jp; Hideyuki Ohnishi - hideyuki@onc.akashi.hyogo.jp



*Corresponding author:

Ryosuke Maeoka,
Department of Neurosurgery,
Nara Medical University,
Kashihara, Nara Prefecture, Japan.
r.maeoka@narmed-u.ac.jp

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ABSTRACT

Background: Superselective shunt occlusion (SSSO) for cavernous sinus dural arteriovenous fistula (CSDAVF) avoids the risk of cranial nerve palsy, unlike entire sinus packing, but requires paying attention to recurrence. Distinguishing between true and paradoxical worsening of postoperative ophthalmic symptoms using a less-invasive modality is often difficult. Here, we report a case of true worsening of neuro-ophthalmic symptom by recurrent CSDAVF detected by venous-arterial spin labeling (ASL) on magnetic resonance imaging.

Case Description: A 55-year-old woman with neither contributory medical history nor previous head trauma presented with neuro-ophthalmic symptoms and pulsatile tinnitus. Digital subtraction angiography (DSA) revealed CSDAVF with multiple shunted pouches. She underwent successful transvenous SSSO, but neuro-ophthalmic symptom worsened after SSSO and venous-ASL revealed increased signal intensity in the right superior orbital vein (SOV). DSA confirmed recurrent CSDAVF and additional transvenous embolization was performed. Neuro-ophthalmic symptoms and venous-ASL hyperintensity on SOV improved postoperatively.

Conclusion: Venous-ASL is noninvasive and seems useful for detecting true worsening of neuro-ophthalmic symptoms of recurrent CSDAVF.

Keywords: Arterial spin labeling, Cavernous sinus dural arteriovenous fistula, Neuro-ophthalmic symptom, Superselective shunt occlusion

INTRODUCTION

Dural arteriovenous fistula (DAVF) is a cerebral vascular malformation characterized by direct communication between dural arteries and either a dural sinus or cortical vein.^[4] Transvenous embolization (TVE) provides effective treatment for cavernous sinus DAVF (CSDAVF). Some reported that superselective shunt occlusion (SSSO) can prevent the risk of permanent cranial nerve palsy caused by the mass effect of coils packing the entire CS.^[1,11,13] However, in cases of CSDAVF with multiple shunts, SSSO could result in residual or recurrent DAVF.^[12] The syndrome of paradoxical worsening after embolization for CSDAVF has been reported as recurrence of neuro-ophthalmic symptoms with clinical improvement of the CSDAVF.^[14] In contrast, true worsening caused by recurrent CSDAVF with increasing blood flow draining into the superior

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orbital vein (SOV) could result in visual impairment. However, estimating whether neuro-ophthalmic symptoms after embolization are true, or paradoxical worsening remains difficult without digital subtraction angiography (DSA). Early detection of true worsening is important. Here, we report a case of recurrent CSDAVF after treatment with SSSO that was detected by venous-arterial spin labeling (ASL).

CASE DESCRIPTION

A 55-year-old woman with neither contributory medical history nor previous head trauma presented with bilateral chemosis, pulsatile tinnitus, exophthalmia, and 6th cranial nerve palsy. Magnetic resonance angiography (MRA) and time-of-flight (TOF) angiography suggested CSDAVF [Figure 1a and b], and cerebral DSA revealed the CSDAVF fed by branches of the internal carotid artery and external carotid artery. DSA also revealed that the CSDAVF had multiple shunted pouches located in bilateral compartments of the CS and draining into bilateral SOVs, inferior petrosal sinuses (IPSs), and superficial middle cerebral veins (SMCVs) [Figure 1c-f]. The patient was diagnosed with CSDAVF, classified as Cognard Grade II a + b

and Barrow type D, and we decided to perform SSSO as TVE. Under general anesthesia, we placed a 6-Fr guiding catheter in the left internal jugular vein through the right femoral vein, in which a 4-Fr catheter had been introduced coaxially with intravenous administration of heparin during endovascular treatment. Heparin had to be given to maintain in the activated clotting time between 250 and 300 s. First, a microcatheter was advanced over the microguidewire into the CS through left IPS, and we performed SSSO of shunt pouches located in the left anteromedial compartment of the CS using coils. TVE was performed to avoid leaving a dangerous drainage route into the superior petrosal sinus and SMCV [Video 1]. Second, SSSO of shunt pouches located in the right anterolateral compartment of the CS by coils was performed through right IPS [Video 2]. Finally, TVE of the drainage route into the right SOV and inter-CS through the left IPS [Video 3]. SSSO was successfully performed [Figure 2a-d]. Immediately after SSSO, the drainage route into the left SOV was seen to be deteriorated, while that into the right SOV only slightly remained. As a result, bilateral neuro-ophthalmic symptoms improved without new cranial nerve palsy and slight remnant right chemosis. No anticoagulant therapy was given postoperatively to promote thrombosis. Postoperative magnetic resonance imaging (MRI)

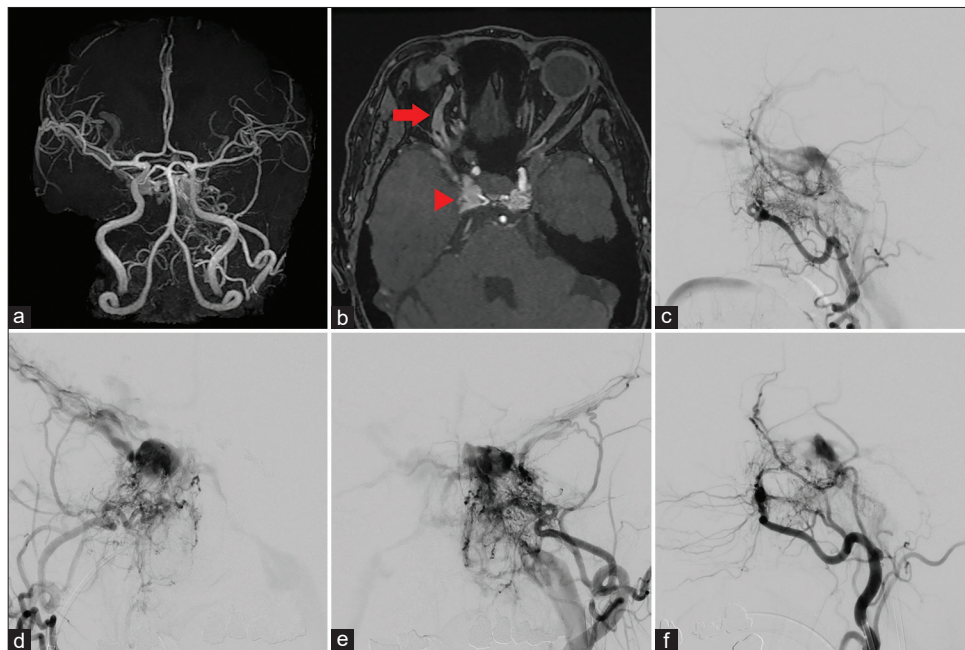


Figure 1: (a) Magnetic resonance angiography suggests cavernous sinus dural arteriovenous fistula (CSDAVF). (b) Time-of-flight angiography suggests CSDAVF (red arrowhead) and dilation of the superior orbital vein (red arrow). (c) Lateral view of right external cerebral angiography reveals the CSDAVF fed by branches of the external carotid artery, has multiple shunted pouches located in the cavernous sinus, and drains into superior orbital vein (SOV), inferior petrosal sinus (IPS), and superficial middle cerebral veins (SMCVs). (d) Anteroposterior (AP) view of right external cerebral angiography reveals the CSDAVF fed by branches of the external carotid artery, has multiple shunted pouches located in the cavernous sinus, and drains into bilateral SOVs, IPSes, and SMCVs. (e) AP view of left external cerebral angiography reveals the CSDAVF fed by branches of the external carotid artery, has multiple shunted pouches located in bilateral compartments of the cavernous sinus, and drains into bilateral SOVs, IPSes, and SMCVs. (f) Lateral view of left external cerebral angiography reveals the CSDAVF fed by branches of the external carotid artery, has multiple shunted pouches located in the cavernous sinus, and drains into SOV, IPS, and SMCVs.

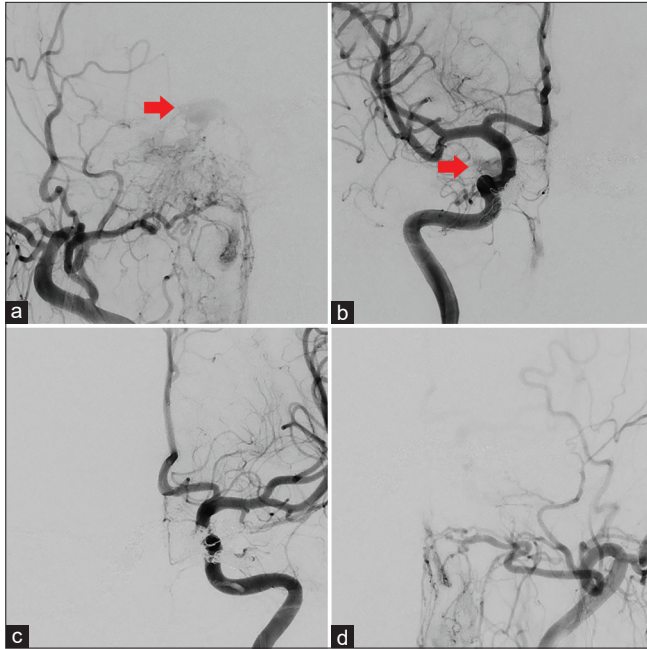


Figure 2: (a) Anteroposterior (AP) view of right external cerebral angiography just after superselective shunt occlusion (SSSO) reveal the drainage route into the left superior orbital vein (SOV) has deteriorated, while that into the right SOV remains to a slight extent (red arrow). (b) AP view of right internal cerebral angiography just after SSSO reveal the drainage route into the left SOV has deteriorated, while that into the right SOV remains to a slight extent (red arrow). (c) AP view of left internal cerebral angiography reveal that the left feeders into the cavernous sinus dural arteriovenous fistula (CSDAVF) have been obliterated. (d) AP view of left external cerebral angiography reveal that the left feeders into the CSDAVF have been obliterated.

showed increased signal intensity in bilateral SOVs on ASL. The imaging parameters of ASL were as follows: Repetition time, 4596 ms; echo time, 10.7 ms; inversion time, 1525 ms; slice thickness, 4 mm; gap, 0 mm; field of view, 240 mm; bandwidth, 62.5 kHz; matrix size, 512 (point) \times 6 (arms); 2 NEX; 30 slice; total acquisition time, 2 min 27 s. ASL images were created using a 3DASL in Functool of Optima MR450w HD (GE Healthcare, Milwaukee, USA) 1.5T for perfusion image analysis. On the other hand, TOF angiography showed dilatation of bilateral SOVs had improved and CSDAVF was obliterated [Figure 3a and b]. One month after SSSO, bilateral neuro-ophthalmic symptoms had worsened, especially on the right side. In contrast, TOF angiography could confirm obliteration of the CSDAVF and improvement of bilateral dilated SOVs. Venous-ASL showed increased signal intensity in bilateral SOVs had improved compared to the one immediately after SSSO, although signal intensity remained slightly in the right SOV [Figure 3c and d]. We considered that bilateral paradoxical worsening of neuro-ophthalmic symptoms had occurred and decided to continue follow-up. However, 2 months after SSSO, only right neuro-ophthalmic symptoms had kept worsening, while those on the left had

resolved. Venous-ASL showed that signal hyperintensity in the right SOV had worsened. Dilatation of only the right SOV and hyperintense signal in the right CS on TOF angiography was also confirmed [Figure 3e and f], and we therefore performed DSA to judge whether true worsening had occurred. DSA revealed recurrence of CSDAVF with a drainage route only into the right SOV [Figure 4a-d]. We performed TVE of the drainage route of the CS into the right SOV via the right IPS [Video 4]. Finally, CSDAVF was obliterated completely on both DSA and venous-ASL [Figure 4e and f]. The postoperative course was uneventful, and neuro-ophthalmic symptoms resolved completely without new cranial nerve palsy.

DISCUSSION

Neuro-ophthalmic symptoms caused by venous reflux into the SOV are common symptoms of CSDAVF and also occur in natural course or after endovascular treatment. The syndrome of paradoxical worsening of neuro-ophthalmic symptoms after endovascular treatment has been attributed to thrombosis of the SOV, the CS, and their tributaries with improvement in the clinical findings of the CSDAVF.^[8,14] In cases of CSDAVF involving multiple shunts, SSSO could result in residual or recurrent DAVF.^[12,13] DSA is the gold standard for diagnosing DAVF because of its superior spatial and temporal resolution. However, DSA is invasive and carries a low but non-negligible risk of neurological and fatal complications and is unsuitable for frequently repeated follow-ups.^[3] Planar computed tomography (CT) and MRI can provide evidence suggestive of DAVF but cannot supply sufficient spatial and temporal resolution of DAVF. Because DAVFs are commonly very small lesions, definitive diagnosis of DAVF requires spatial and temporal resolution comparable to those of DSA. Four-dimensional CT angiography and time-resolved contrast-enhanced MRA are less invasive than DSA and are considered effective modalities for the diagnosis of DAVF,^[7,15] but risk complications induced by contrast agents. In contrast, venous-ASL is now widely available on clinical MRI scanners and utilizes water protons in arterial blood as endogenous tracers to assess cerebral blood flow (CBF), providing a repeatable MR perfusion method for measuring CBF quantitatively without gadolinium.^[5,6] Increased local blood flow has been demonstrated as signal hyperintensity on ASL. Identification of an increased signal intensity on venous-ASL improves the diagnostic accuracy and increases confidence in the presence or absence of DAVF in a noninvasive manner. As a result, venous-ASL offers high sensitivity and specificity for the presence of DAVF, particularly CSDAVF, and may help minimize exposure to the risks of diagnostic DSA.^[2] The sensitivity and specificity of venous ASL single intensity to predict a positive DSA study were 78% and 85%, respectively.^[10] TOF-MRA is also available on clinical MRI scanners and is a completely noninvasive

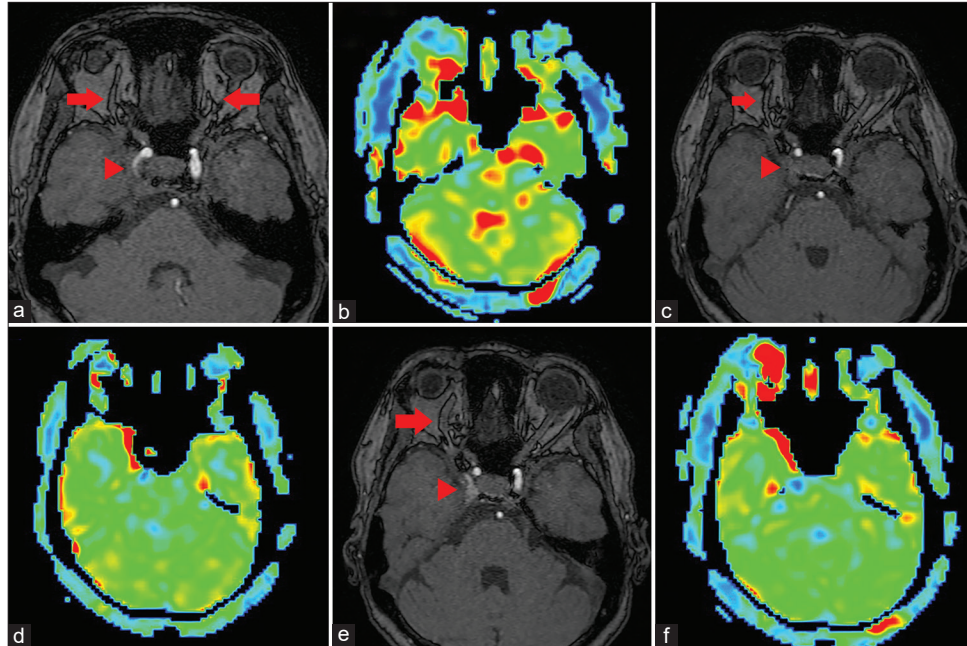


Figure 3: (a) Time-of-flight (TOF) angiography just after first operation shows dilatation of bilateral superior orbital veins (SOVs) has improved (red arrow) and the cavernous sinus dural arteriovenous fistula (CSDAVF) has been obliterated (red arrowhead). (b) Venous-arterial spin labeling (ASL) confirms increased signal intensity at bilateral SOVs. (c) TOF angiography on the 1 month later from superselective shunt occlusion (SSSO) shows dilatation of the SOVs (red arrow) and obliteration of the CSDAVF (red arrowhead). (d) Venous-ASL on the 1 month later from SSSO reveals resolution of increased signal intensity in the left SOV, but slight remnant hyperintensity of the right SOV. (e) TOF angiography on the 2 months later from SSSO shows dilatation of only the right SOV (red arrow) and confirmation of hyperintense signal in the right CS (red arrowhead). (f) Venous-ASL on the 2 months later from SSSO shows exacerbation of signal hyperintensity in the right SOV.

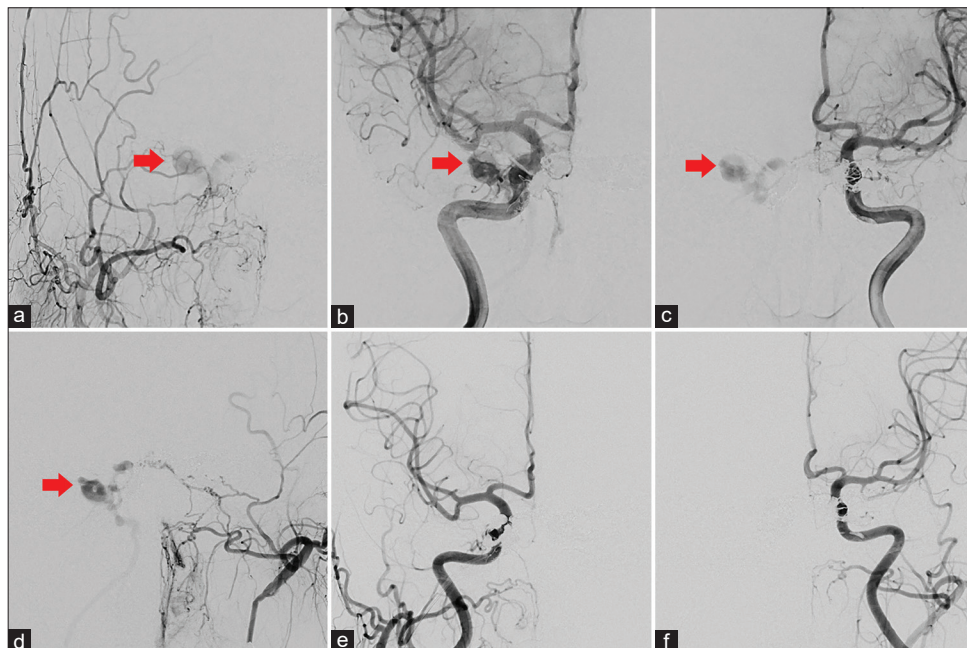


Figure 4: (a) Right external cerebral angiography demonstrate recurrence of cavernous sinus dural arteriovenous fistula (CSDAVF) draining into the right superior orbital vein (SOV) (red arrow). (b) Right internal cerebral angiography demonstrate recurrence of CSDAVF draining into the right SOV (red arrow). (c) Left internal cerebral angiography also demonstrate recurrence of CSDAVF draining into the right SOV (red arrow). (d) Left external cerebral angiography also demonstrate recurrence of CSDAVF draining into the right SOV (red arrow). (e) Postoperative right cerebral angiography reveals complete obliteration of the CSDAVF. (f) Postoperative left cerebral angiography reveals complete obliteration of the CSDAVF.

modality, but offers lower sensitivity than venous-ASL for the presence of DAVF.^[2,9] In the present case, bilateral neuro-ophthalmic symptoms were worsened at 1 month after from SSSO. However, increased signal intensity in the left SOV had completely resolved and that in the right SOV remained to a slight extent despite the fact that TOF-MRA showed CSDAVF had been completely obliterated. Finally, DSA revealed recurrence of CSDAVF with a drainage route into only the right SOV at 2 months after SSSO. Venous-ASL seems useful for detecting true worsening of CSDAVF. If venous-ASL demonstrates increased signal intensity in the SOV in a patient suffering from CSDAVF, DSA may be warranted.

We recognize that venous-ASL has some drawbacks. First, artifacts may be present in the image. False-positives and false-negatives can occur, and the sensitivity of venous-ASL is inferior to that of DSA. Second, ASL offered limited spatial resolution and the classification of DAVF could not be determined from ASL alone. Definitive diagnosis of DAVF thus requires DSA, as does planning of the treatment strategy.

CONCLUSION

We have reported a case of recurrent CSDAVF after treatment with SSSO, detected by venous-ASL on MRI. Venous-ASL offers high sensitivity and specificity for the presence of DAVF and seems useful for completely noninvasive differentiation between true and paradoxical worsening of neuro-ophthalmic symptoms of CSDAVF.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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