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Case Report

Arterial Spin Labeling Was Useful for Evaluating the Treatment Response of a Transverse–Sigmoid Sinus Dural Arteriovenous Fistula: A Case Report

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Objective: We report the case of a patient in whom arterial spin labeling (ASL) was useful for assessing the effects of treatment for a transverse–sigmoid sinus dural arteriovenous fistula (TSS-dAVF).

Case Presentation: The patient was a 65-year-old man. Cerebral angiography demonstrated an aggressive dAVF involving the TSS, superior sagittal sinus (SSS), and the sinus confluence, with severe cortical and deep venous reflux. We performed multiple transarterial and transvenous embolizations for the TSS and sinus confluence lesion. The shunt disappeared almost completely after embolization. A high signal intensity that had been apparent in the SSS and straight sinus (StS) on ASL imaging before embolization disappeared after embolization. ASL imaging 3 months after embolization revealed slightly a high signal intensity in the StS, which was considered to be due to recurrence of the lesion. Moreover, recurrence of the confluence and TSS-dAVF was observed on cerebral angiography 6 months after embolization. As additional embolization was considered difficult, radiation therapy was recommended, but the patient refused; therefore, follow-up was performed. As ASL imaging findings were consistent with cerebral angiography findings, careful examination and monitoring of changes on ASL imaging were subsequently performed.

Conclusion: Follow-up using ASL imaging is useful to assess the effects of treatment performed for a dAVF.

Keywords▶ dural arteriovenous fistula, arterial spin labeling

Introduction

Arterial spin labeling (ASL) is an imaging technique to obtain non-contrast-enhanced perfusion images using an MRI system. It is routinely applied for the diagnosis of cerebrovascular disorder[1)] or to assess the malignancy of brain tumors[2)] in clinical practice. In the diagnosis of dural arteriovenous fistula (dAVF), cerebral angiography is the gold standard, but ASL facilitates the assessment of sinus or cortical vein outflow routes and can be utilized for cortical venous reflux (CVR) assessment.[3)] In this study, we report a patient in whom ASL was useful for evaluating the treatment response of a transverse–sigmoid sinus (TSS)-dAVF and review the literature.

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Case Presentation

Patient: The patient was a 65-year-old man.
Complaint: The patient complained of amnesia.
Medical history: The patient had a medical history of hypertension, dyslipidemia, and prostatic hypertrophy.
Family history: The patient's family history was not contributory.
Present illness: The patient had noticed amnesia for the past month. The symptom had gradually exacerbated. He had a car accident the day before admission and was instructed not to drive a car. However, he went to work on a farm by a car on the day of arrival. Subsequently, weakness of the lower limbs was noted and he repeatedly fell

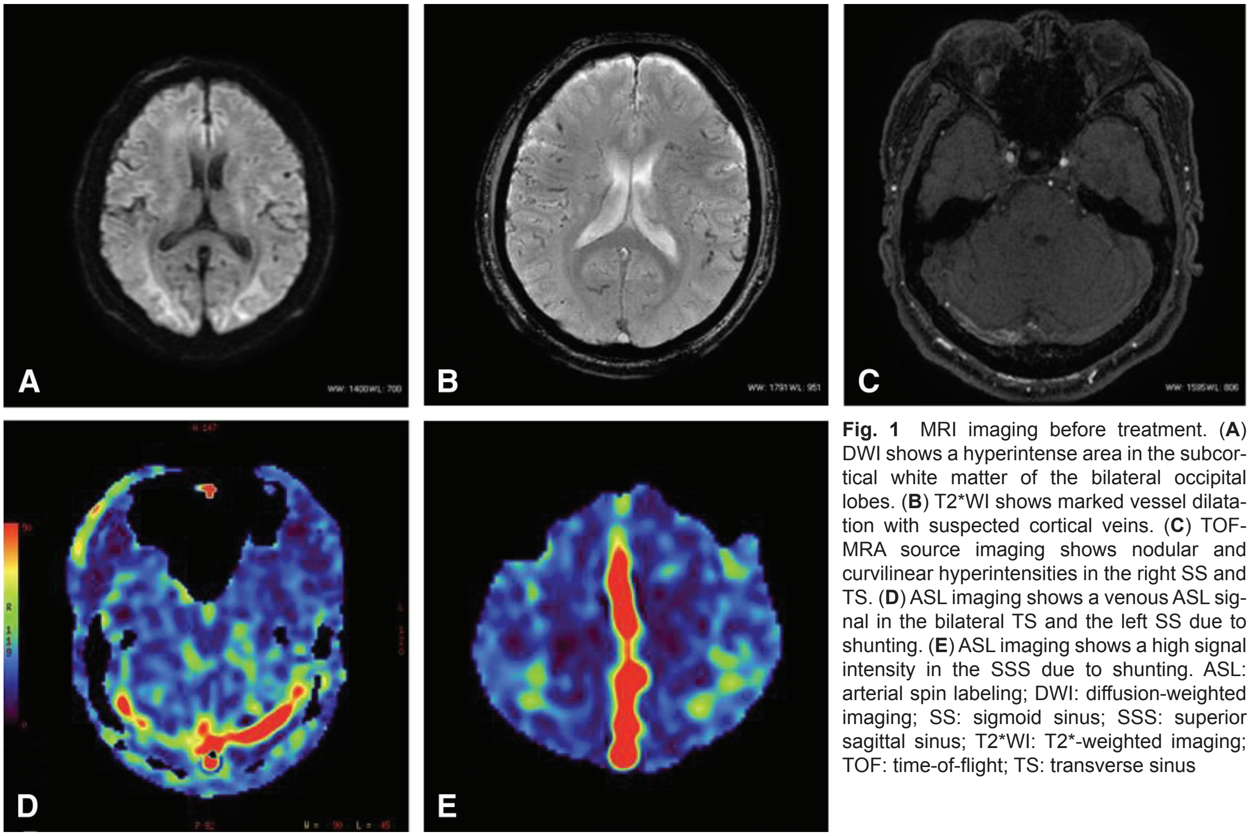

Fig. 1 MRI imaging before treatment. (**A**) DWI shows a hyperintense area in the subcortical white matter of the bilateral occipital lobes. (**B**) T2*WI shows marked vessel dilatation with suspected cortical veins. (**C**) TOF-MRA source imaging shows nodular and curvilinear hyperintensities in the right SS and TS. (**D**) ASL imaging shows a venous ASL signal in the bilateral TS and the left SS due to shunting. (**E**) ASL imaging shows a high signal intensity in the SSS due to shunting. ASL: arterial spin labeling; DWI: diffusion-weighted imaging; SS: sigmoid sinus; SSS: superior sagittal sinus; T2*WI: T2*-weighted imaging; TOF: time-of-flight; TS: transverse sinus

down. He was found lying down in a tunnel, and was brought to the Emergency and Critical Care Center of our hospital by ambulance.

Physical examination on admission

The Japan Coma Scale score was 1. The Glasgow Coma Scale score was 15 (E4V5M6). There was no paralysis of the limbs, but cognitive impairment was suspected.

Imaging findings

Diffusion-weighted imaging (DWI) demonstrated a high signal intensity below the cortexes of the bilateral occipital lobes (**Fig. 1A**). On an apparent diffusion coefficient (ADC) map, there was a slight increase at the same site, suggesting vasogenic edema related to venous congestion (not presented). T2-weighted imaging (T2WI) revealed marked dilation of the cortical veins (**Fig. 1B**). Linear/punctiform high signal intensity involving the right sigmoid sinus (SS), transverse sinus (TS), and sinus confluence was observed on the time-of-flight (TOF)-MRA source imaging, suggesting multiple shunts from the bilateral occipital arteries (OAs) (**Fig. 1C**). ASL revealed hyperperfusion to the right TS, left TS to SS, superior sagittal sinus (SSS), and straight sinus (StS) (**Fig. 1D** and **1E**).

Cerebral angiography confirmed diffuse shunts of the right TS to SS, partially involving the sinus confluence, from the bilateral OAs, middle meningeal arteries, right posterior cerebral artery (PCA), posterior meningeal artery, and left superior cerebellar artery as feeders. Localized stenosis of the left TS to SS, in addition to markedly extensive stenosis of the right TS to SS, was also observed. Retrograde venous reflux from the SSS and StS to the cortical and deep veins was noted (**Fig. 2A–2C**). Cerebral/cerebellarvenous congestion was marked, and a pseudophlebitic pattern was observed (**Fig. 2D**).

Treatment course

Shunts from several arteries were present, and we selected a strategy to reduce shunt blood flow using transarterial embolization (TAE) and transvenous embolization (TVE). In the first session of TAE, the dural branch of right OAs was involved and was selectively embolized with 10% *n*-butyl-2-cyanoacrylate (NBCA). In the second session, the dural branch of left OAs was involved and selectively embolized with 10% NBCA. Although disappearance of the shunts was not achieved, shunt blood flow was markedly reduced. In the third session, a 4F long sheath was inserted into the left femoral artery and a 6F long sheath was inserted into the right

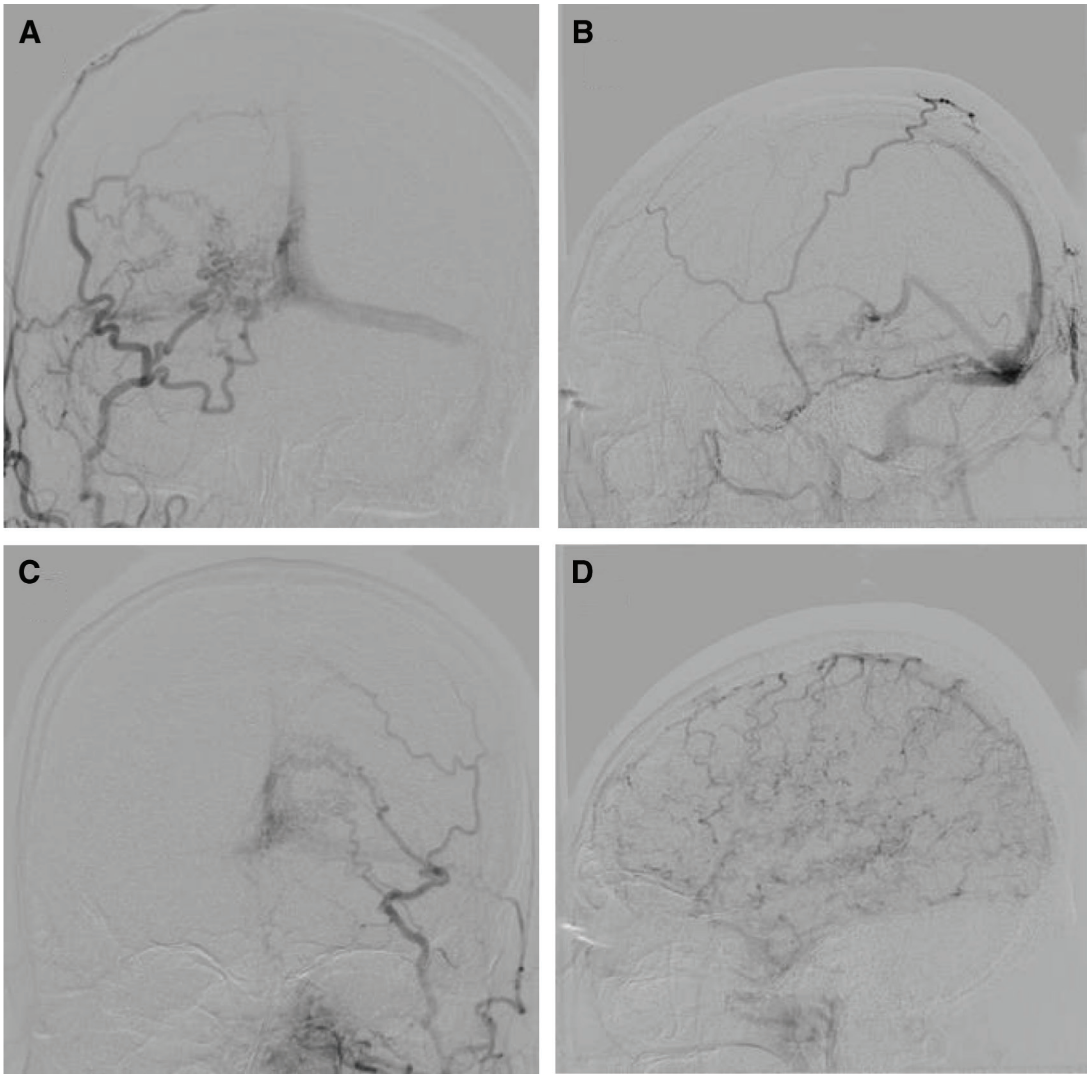

Fig. 2 Cerebral angiography imaging before treatment. (**A**) Right external carotid angiography (anteroposterior view) shows a TSS-dAVF supplied by the right OA and MMA. (**B**) Right external carotid angiography (lateral view) shows a high-flow arteriovenous shunt to the sinus, and retrograde sinus drainage to the superior sagittal and StSs. (**C**) Left external carotid angiography (anteroposterior view) shows a TSS-dAVF supplied by the left OA and MMA. (**D**) Right internal carotid angiography (lateral view) shows cortical vein dilatation in the cerebrum and cerebellum with a PPP. MMA: middle meningeal artery; OA: occipital artery; PPP: pseudophlebitic pattern; StS: straight sinus; TSS-dAVF: transverse–sigmoid sinus dural arteriovenous fistula

femoral vein. Systemic heparinization was performed. A 6F ENVOY (Johnson & Johnson, New Brunswick, NJ, USA) was inserted into the left internal jugular vein, and a Cerulean G 127 cm (Medikit, Tokyo, Japan) was inserted into the distal left TS via the left SS to TS. An Echelon-14 (Medtronic, Irvine, CA, USA) was inserted to the site of the distal right TS shunt using a Synchro2 soft (Stryker, Kalamazoo, MI, USA). Sinus packing was conducted using a total of 27 coils (186 cm) such as a Target XL 3 mm × 9 cm (Stryker), HyperSoft 3 mm × 8 cm (Terumo, Tokyo, Japan), and SMART COIL 3 mm × 8 cm (Penumbra, Alameda, CA, USA). Subsequently, an ASAHI CHIKAI 315 EXC (Asahi Intecc, Aichi, Japan) was inserted into the SSS, and the left TS to SS stenotic site was dilated at 8 atm for 30 sec using an Aviator Plus 5 mm × 40 mm (Johnson & Johnson). Regurgitation to the SSS and deep vein decreased, and anterograde blood flow from the left TS to SS increased. In the fourth session, a shunt remained at the venous pouch of the proximal inferior right transverse/sigmoid sinuses involving the bilateral OA branches and cortical branch at the periphery of the right PCA as feeders, and it was embolized using a total of 6 detachable coils (27 cm), as described for the previous session. Bilateral external carotid angiography confirmed disappearance of the shunt (**Fig. 3A** and **3B**).

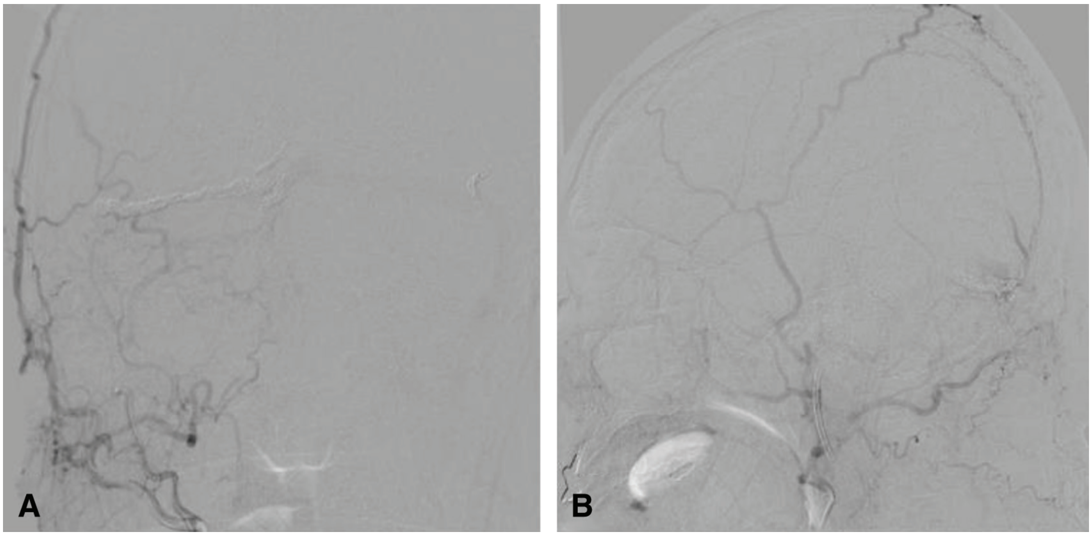

Fig. 3 Cerebral angiography imaging immediately after embolization. (**A**) Right external carotid angiography after embolization (anteroposterior view) confirmed the disappearance of the shunt supplied by the right OA and MMA disappeared. (**B**) Right external carotid angiography after embolization (lateral view) confirmed the disappearance of the retrograde sinus drainage to the superior sagittal and StSs. MMA: middle meningeal artery; OA: occipital artery; StS: straight sinus

Postoperative course

A slight shunt flow remained at the sinus confluence, but it was markedly reduced. A TOF-MRA source image 5 days after surgery demonstrated slight, punctiform high signal intensity at the sinus confluence. ASL revealed hyperperfusion at the same site (**Fig. 4A** and **4B**). However, there was no new neurological deficit and the patient was discharged 47 days after admission. Three months after surgery, an increase in the high-signal-intensity area at the sinus confluence was noted on TOF-MRA and ASL suggested enlargement of the hyperperfusion area at the sinus confluence to the left TS (**Fig. 4C** and **4D**). We considered two possibilities: the shunt may have increased or restenosis of the sinus after sinus plasty may have occurred, increasing sinus regurgitation. DWI confirmed the disappearance of the high signal intensity below the cortexes of the bilateral occipital lobes. T2WI also confirmed the disappearance of the dilated vessel (**Fig. 4E** and **4F**). Cerebral angiography 6 months after surgery did not reveal restenosis of the sinus after sinus plasty. There was an increase in the number of shunts at areas adjacent to the right TS and sinus confluence (**Fig. 5A** and **5B**). TVE was attempted, but approaching was impossible and additional treatment was abandoned. Radiotherapy was recommended, but the patient requested follow-up. On ASL 1 year and 3 months after surgery, hyperperfusion at the StS was observed, suggesting retrograde blood flow, as noted on cerebral angiography. Careful follow-up was continued while paying attention to changes in the high-signal-intensity area on TOF-MRA source images and hyperperfusion area on ASL (**Fig. 6A–6H**).

Discussion

Kuwayama et al. previously reported the incidence of dAVF in Japan to be 0.29 per 100,000 persons.[4] This disorder is relatively rare. Regarding sites, its incidences at the cavernous sinus and TSS are 43.6% and 33.4%, respectively, according to the Japanese Registry of Neuroendovascular Therapy.[5]

Concerning the indications of treatment, aggressive therapeutic intervention is required for dAVF with CVR. Satomi et al. found no symptom deterioration in 98.2% of dAVF patients without CVR, whereas there was a change to an aggressive pattern in approximately 2% of such patients.[6] Therefore, careful follow-up may be necessary even in CVR-free patients. Furthermore, van Dijk et al. reported that the annual mortality rate among dAVF patients with CVR was 10% and that the annual incidence of serious adverse events related to central nervous system disorder was 15%.[7] Therefore, aggressive treatment is indicated for patients with cerebral venous congestion due to retrograde sinus reflux or CVR. In many cases in which the shunt blood flow volume is large despite a major lesion of the TSSs and a portion of the shunt is present at the sinus confluence, as demonstrated in the present case, radical cure by complete shunt disappearance is difficult. Many dAVF at the sinus confluence are aggressive, with a complete occlusion rate of 25%, and

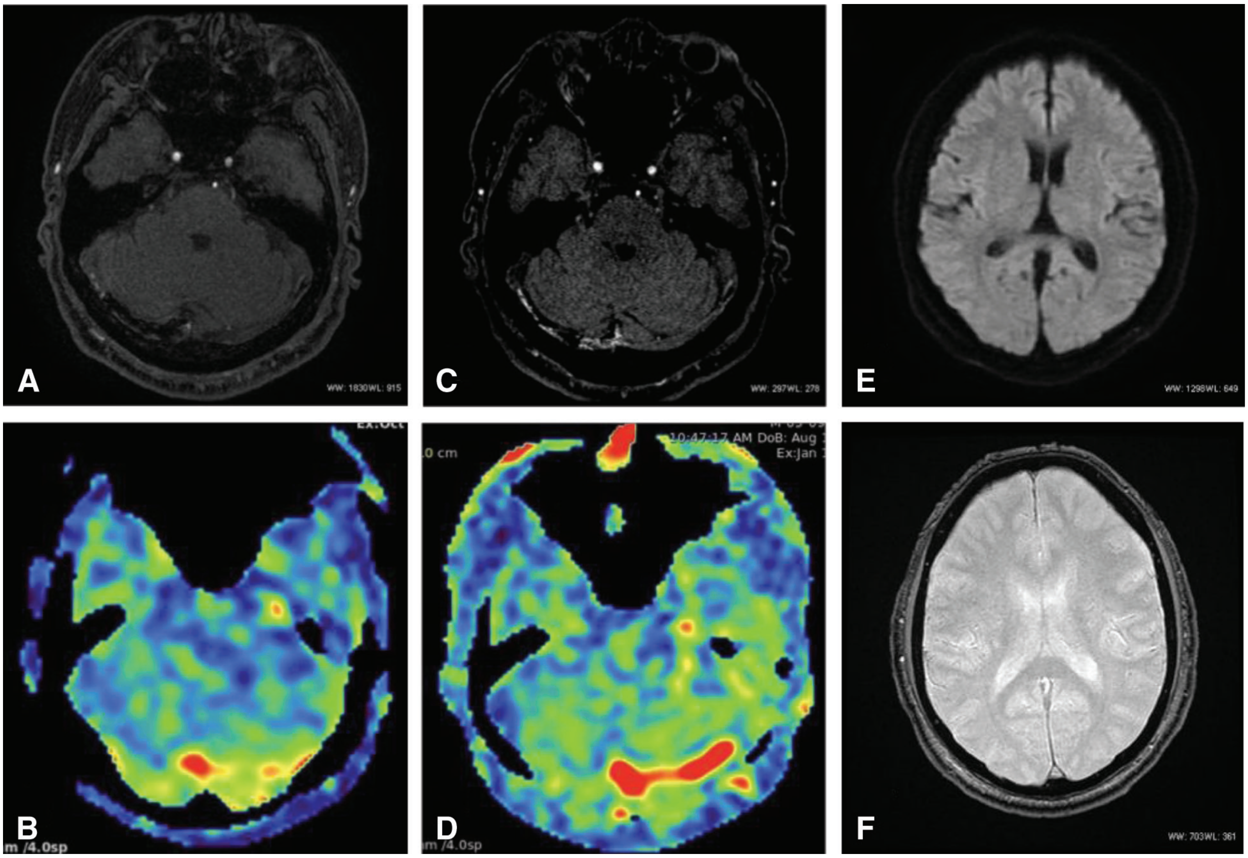

Fig. 4 MRI imaging after embolization: 5 days after embolization and 3 months after embolization. (**A**) TOF-MRA source imaging 5 days after embolization shows the slight hyperintensities in the sinus confluence. (**B**) ASL imaging 5 days after embolization shows a high signal intensity in the sinus confluence. (**C**) TOF-MRA source imaging 3 months after embolization shows the increased hyperintensities in the sinus confluence. (**D**) ASL imaging 3 months after embolization shows a high signal intensity in the sinus confluence and left TS. (**E**) DWI 3 months after embolization shows the disappearance of the hyperintense area in the bilateral occipital lobes. (**F**) T2*WI 3 months after embolization shows the disappearance of the vessel dilatation with suspected cortical veins. ASL: arterial spin labeling; DWI: diffusion-weighted imaging; T2*WI: T2*-weighted imaging; TOF: time-of-flight; TS: transverse sinus

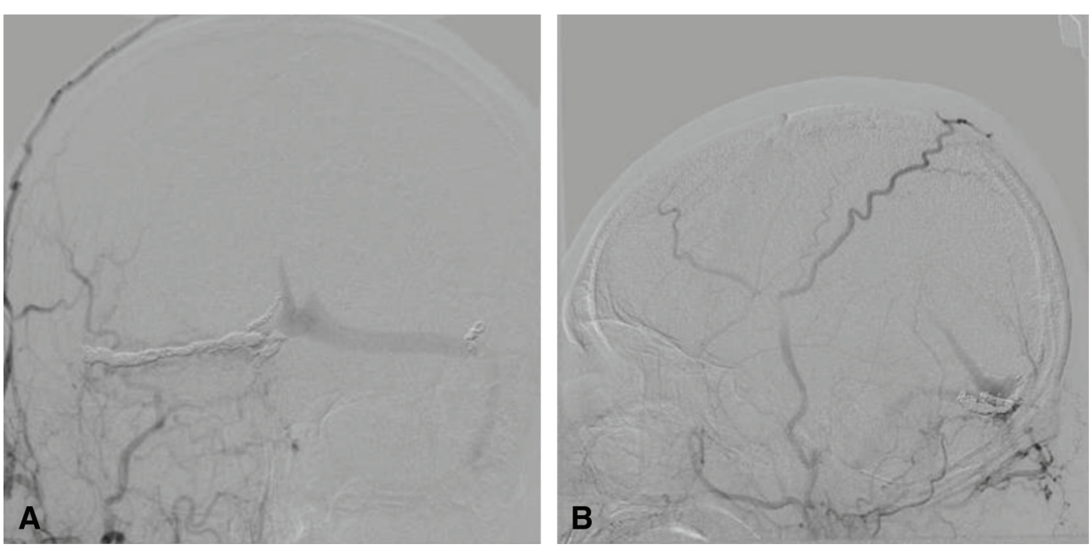

Fig. 5 Cerebral angiography imaging 6 months after embolization. (**A**) Right external carotid angiography 6 months after embolization (anteroposterior view) shows the recurrence of the confluence and TSS-dAVF supplied by the right OA and PAA. (**B**) Right external carotid angiography 6 months after embolization (lateral view) shows the presence of retrograde drainage to the StS. OA: occipital artery; PAA: posterior auricular artery; TSS-dAVF: transverse–sigmoid sinus dural arteriovenous fistula; StS: straight sinus

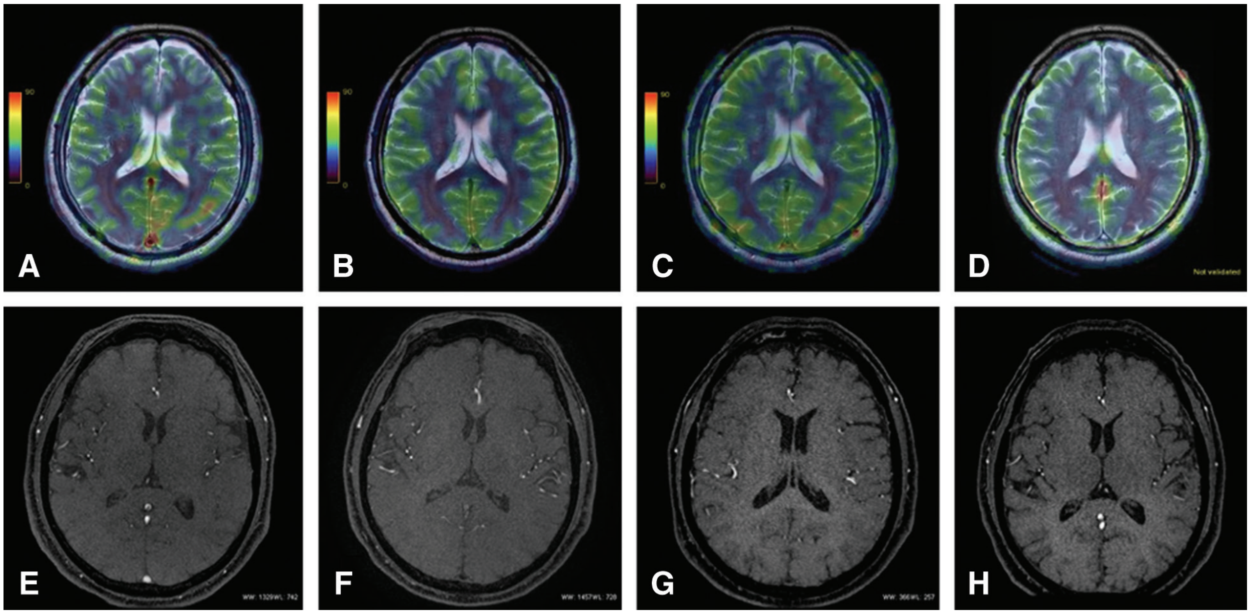

Fig. 6 ASL color map and T2 fusion imaging, TOF-MRA source imaging of treatment progress. (**A**) ASL color map and T2 fusion imaging before embolization show a high signal intensity in the SSS and StS due to shunting. (**B**) ASL color map and T2 fusion imaging 5 days after embolization show the disappearance of the high signal intensity in the SSS and StS. (**C**) ASL color map and T2 fusion imaging 3 months after embolization show the recurrence of slightly high signal intensity in the StS. (**D**) ASL color map and T2 fusion imaging a year after embolization show the recurrence of markedly high signal intensity in the StS. (**E**) TOF-MRA source imaging before embolization shows hyperintensities in the SSS and StS due to shunting. (**F**) TOF-MRA source imaging 5 days after embolization shows the slight hyperintensities in the StS. (**G**) TOF-MRA source imaging 3 months after embolization shows the disappearance of hyperintensities in the StS. (**H**) TOF-MRA source imaging a year after embolization shows the recurrence of marked hyperintensities in the StS. ASL: arterial spin labeling; SSS: superior sagittal sinus; StS: straight sinus; TOF: time-of-flight

radical cure is difficult.[8] Therefore, posttreatment follow-up imaging is important in cases in which complete occlusion was not achieved such as the present case.

Recently, advances in diagnostic devices have facilitated noninvasive diagnosis/follow-up. TOF-MRA source images and ASL are useful.[9] On the former, innumerable high signal intensities are visualized in the dilated meningeal artery or in/around the sinus wall, and are useful for the diagnosis of shunt points.[10] Furthermore, ASL is a method in which arterial blood flowing in the brain tissue is labeled with radio frequency (RF) pulses in the absence of contrast medium, and is used as an intrinsic tracer for noninvasive cerebral perfusion imaging. It facilitates the detection of outflow veins, and is useful for pre-/postoperative assessment, follow-up, and screening. On ASL, dAVF shunt sites or outflow veins exhibit hyperperfusion and can be visually recognized.[11] This procedure is minimally invasive, differing from cerebral angiography. Radiation exposure-free examination can be performed. The invasiveness of contrast-enhanced MRI, such as MR-DSA, is also relatively low, and this procedure is useful. However, a previous study reported the intracerebral retention of gadolinium,[12] although it may not induce clinically significant symptoms. Furthermore, serious adverse reactions, such as nephrogenic systemic fibrosis (NSF) and anaphylactic shock, may develop. In patients with renal dysfunction or bronchial asthma, the use of gadolinium contrast medium is sometimes difficult; follow-up using non-contrast-enhanced imaging procedures, such as TOF-MRA and ASL, may be clinically valuable. As a caution for ASL, a waiting time until the labeled blood is perfused to the brain tissue, known as post-label delay (PLD), is present. Concerning the optimal timing of PLD, a study involving healthy adults found that optimal images can be obtained at a PLD of 1500 to 2000 msec.[13] Furthermore, another study reported the influence of age; ASL-cerebral blood flow decreased with age in comparison with young persons.[14] In our hospital, a PLD of 2025 msec is adopted as a criterion, but there are individual differences in the interval until the labeled blood reaches the region of interest; therefore, regarding a sufficient PLD, comparison with gold standards to directly evaluate an increase or decrease in shunt blood flow, such as cerebral angiography and 4D CT/MR imaging, in addition to case accumulation in the future, is necessary.

We compared the properties of non-contrast-enhanced images. As the advantages of TOF-MRA, a high-flow site is detected as a signal through in-flow effects, and the inflow artery to the shunt site is visualized as the highest signal intensity; findings similar to those on cerebral angiography can be noninvasively evaluated. As its limitations, signals are weak and evaluation using maximum intensity projection (MIP) or volume rendering (VR) rearrangement images is difficult; therefore, original images must be carefully evaluated. In addition, findings may not be recognized as significant in the case of a fine shunt. As the advantages of ASL, outflow vein blood flow is visualized as a high signal intensity; the sinus with regurgitation and CVR can be sensitively evaluated. In addition, a color mapping display facilitates visual recognition. As its limitations, hyperperfusion is sometimes observed even in a significance-free area in healthy adults; interpretation is difficult in some patients. In addition, when the PLD is insufficient, the lesion may be overlooked.

In the present case, we performed several treatment sessions for a TSS-dAVF, and confirmed blood flow changes using ASL. The disappearance or recurrence of a shunt on angiography was also reflected by ASL. In particular, careful follow-up is necessary when complete embolization is not achieved. However, repeated angiography has risks, and follow-up by MRI, including ASL, may be useful.

Conclusion

We reported a patient in whom ASL was useful for evaluating the treatment response of a TSS-dAVF. The disappearance or recurrence of a shunt on angiography was also reflected by ASL. ASL, as an additional procedure, may be useful for evaluating the treatment response.

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Disclosure Statement

The authors declare no conflict of interest.

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