Radiology Case Reports 16 (2021) 1655–1659

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case report

Usefulness of arterial spin labeling in the evaluation for dural arteriovenous fistula of the craniocervical junction☆

Seishiro Takamatsu, M.D.[a,b,c,*], *Kohei Suzuki, M.D.*[a], *Yu Murakami, M.D., Ph.D*[d], *Kei Nomura, M.D., Ph.D*[c], *Junkoh Yamamoto, M.D., Ph.D*[a], *Shigeru Nishizawa, M.D., Ph.D*[a]

[a] *Department of Neurosurgery, University of Occupational and Environmental Health, Kitakyushu, Fukuoka, Japan*
[b] *Seirei Center for Health Promotion and Preventive Medicine, Hamamatsu, Shizuoka, Japan*
[c] *Center for Brain and Spine Surgery, Aoyama General Hospital, Toyokawa, Aichi, Japan*
[d] *Department of Radiology, University of Occupational and Environmental Health, Kitakyushu, Fukuoka, Japan*

ARTICLE INFO

Article history:
Received 26 January 2021
Revised 2 April 2021
Accepted 2 April 2021

Keywords:
Arterial spin labeling
Dural arteriovenous fistula
Craniocervical junction

ABSTRACT

In the diagnosis of an intracranial dural arteriovenous fistula (DAVF), arterial spin labeling (ASL), a sequence of magnetic resonance imaging (MRI) to depict high-blood-flow intracranial lesions, has been reported as a useful and noninvasive tool, not only to predict the presence of cortical venous drainage and draining veins, but also to confirm persistent obliteration after treatment. However, such utility of ASL has not been reported in DAVF of the craniocervical junction (CCJDAVF) because of the rarity of this disease and uncertainty in the acquisition of precise images.

We report a case of CCJDAVF presenting with myelopathy. Preoperative ASL images showed an abnormal high-intensity signal in the craniocervical junction, consistent with the anterior spinal vein and draining veins, which were also identified by digital subtraction angiography. After successful surgical treatment for the disease, MRI and 4-dimensional computed tomography angiography (4DCTA) confirmed complete disappearance of CCJDAVF. The ASL images also showed no abnormal intensity signal. The patient was followed-up using ASL, and no recurrence of high-intensity signal was observed.

As repetitive image examination is mandatory in the follow-up of a patient with DAVF to exclude recurrence, ASL is highly beneficial because of the unnecessity of an exogenous contrast medium and high credibility to depict the disease. The craniocervical junction may

☆ Competing interest: The authors report no conflicts of interest related to the materials or methods used in this study or the findings specified in this paper.

* Corresponding author: Seishiro Takamatsu, Department of Neurosurgery, University of Occupational and Environmental Health, 1-1 Iseigaoka, Yahatanishi-ku, Kitakyushu, Fukuoka 807-8555, JAPAN.
E-mail address: stakamatsu617@gmail.com (S. Takamatsu).

https://doi.org/10.1016/j.radcr.2021.04.006

be out of the field of view in routine MRI. Special attention must be paid to setting the field of view and post labeling delay (PLD) to obtain precise images of ASL in CCJDAVF.

Introduction

A Dural arteriovenous fistula (DAVF) can arise at any portion of the dura mater. The dilated veins and fistulas of DAVF might cause intracerebral hemorrhage and/or subarachnoid hemorrhage. To correctly diagnose such lesions, to select an appropriate treatment, and to follow-up, it is no doubt that digital subtraction angiography (DSA) is an essential tool for it. However, it is also true that less frequent examinations using DSA are profitable for patients.

Arterial spin labeling (ASL), a sequence of magnetic resonance imaging (MRI), is a noninvasive neuroimaging technique used to obtain information on cerebral blood flow (CBF) by utilizing electromagnetically labeled arterial blood as an endogenous tracer instead of an exogenous tracer such as a contrast medium. [1] In combination with other modalities, ASL is sensitive for hyperperfusion lesions such as stroke, tumors, and seizures. [2,3,4] In addition, the usefulness of ASL for DAVF diagnosis has also been described in the literature, and the possibility of the prediction of cortical venous drainage in the initial stage of diagnosis and confirming persistent obliteration after treatment using ASL have been discussed.[5,6,7,8] Pre-and postoperative repetitive neuroimaging examinations are mandatory for patients with DAVF; a noninvasive tool will be highly beneficial for patients if it can correctly depict the high-flow disease.

The DAVF of the craniocervical junction (CCJDAVF) is one of the DAVFs, which is an extremely rare disease and is very difficult to diagnose accurately and treat. There have been no reports on diagnosis of CCJDAVF and follow-up using ASL in CCJDAVF because of the rarity of the disease. Therefore, it is unclear whether ASL would be as useful for CCJDAVF as it is for other intracranial DAVFs. Hence, we report a patient with CCJDAVF who was examined by ASL before and after treatment and discuss the clinical significance of ASL in CCJDAVF.

Case report

A 68-year-old woman presented with numbness in bilateral lower extremities. As her numbness gradually extended to the bilateral upper extremities, and she felt the motor weakness of the bilateral upper extremities and was unable to be ambulatory five months later, she visited our hospital.

Neurological examination revealed severe hypoesthesia of the bilateral lower extremities and weakness in all extremities.

The patient was examined with a 3T MRI system (Discovery MR750w; GE Healthcare, Milwaukee, WI, USA) using a dedicated 8-channel phased array coil (Invivo, Gainesville, Florida).

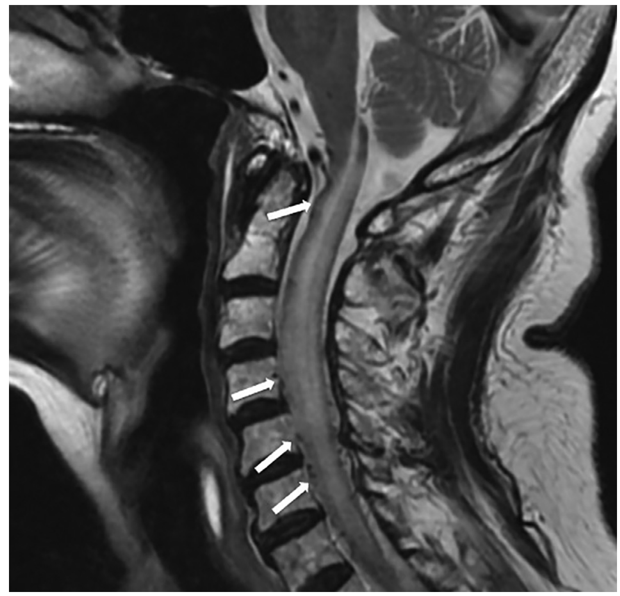

Fig. 1 – The preoperative MRI T2 weighted sagittal image showing swelling of the spinal cord. Abnormal flow void signals (arrows) were detected in the subdural space of the ventral cervical cord. MRI, magnetic resonance imaging.

The ASL imaging was performed using a pseudo-continuous ASL pulse sequence. The ASL parameters were as follows: labeling pulse duration = 1.5s, post labeling delay(PLD) = 1.5 and 2.5s, TR=4446-4564 ms, TE= 9.4-9.9ms, field of view (FOV) = 240 × 240mm, NEX = 2, number of interleaved slices = 30, and slice thickness = 4 mm. The MRI showed swelling of the medulla oblongata and the spinal cord until the upper thoracic levels, and an intramedullary high-intensity area was seen on T2-weighted images. Abnormal flow voids were detected in the subdural space of the ventral cervical cord. (Fig. 1, arrows) The ASL images revealed an apparent high-intensity signal on the ventral side of the brainstem especially in PLD = 1.5s. (Fig. 2A arrow) The DSA and 3-dimensional digital subtraction angiography of the vertebral artery (VA) revealed a CCJDAVF, which was fed by the right meningeal arteries from the right VA that drained into the dilated anterior spinal vein (ASV). (Figs. 3A and B) arrows and arrowheads). In addition, axial view of 4-dimensional computed tomography angiography (4DCTA) demonstrated fistula points around the site of the dural penetration of the right VA. (Fig. 3C arrows) In all together, a high-intensity signal in the ASL images was recognized as dilated ASV.

As the feeding arteries were thin and so bending in the DSA findings, selective catheterization into the feeing arteries and injection of embolic material were considered to be difficult and might cause spinal/brainstem infarction. There-

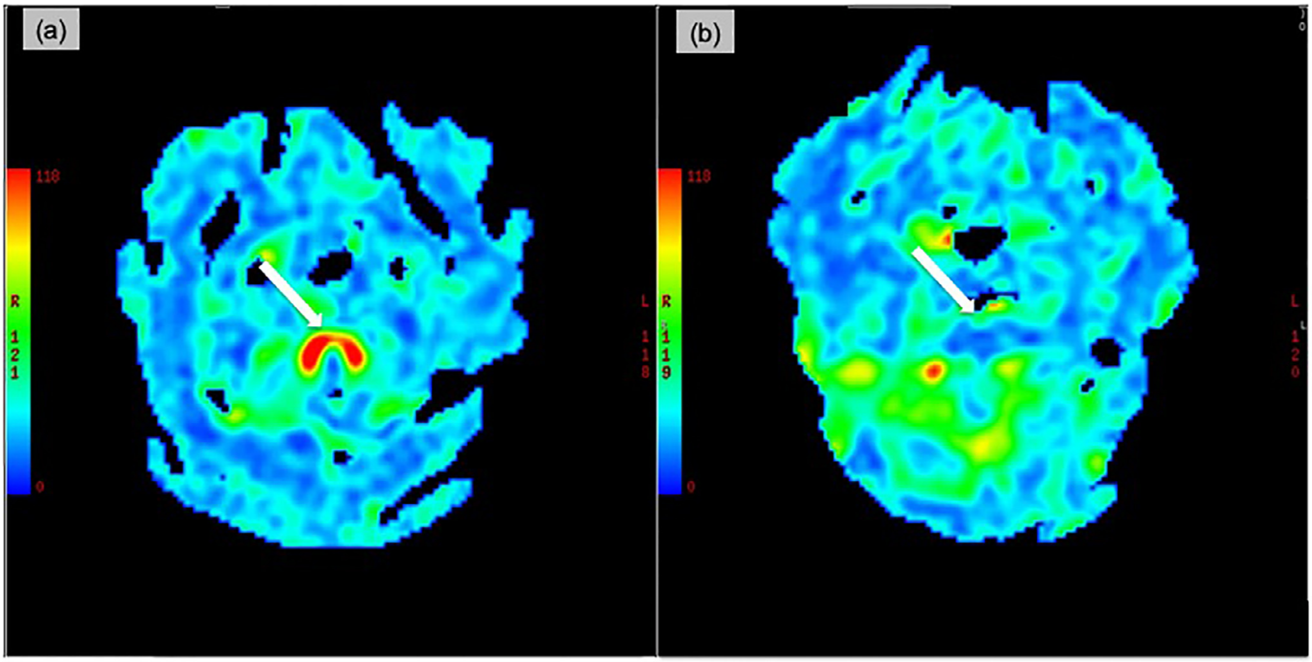

Fig. 2 – The ASL images at the level of medulla oblongata (PLD = 1.5s). The preoperative ASL shows visible high-intensity signal in the ventral medulla oblongata (Fig 2, A, arrow); such an intense signal lesion is not seen in the postoperative ASL (Fig 2, B, arrow). ASL, arterial spin labeling; PLD, post labeling delay.

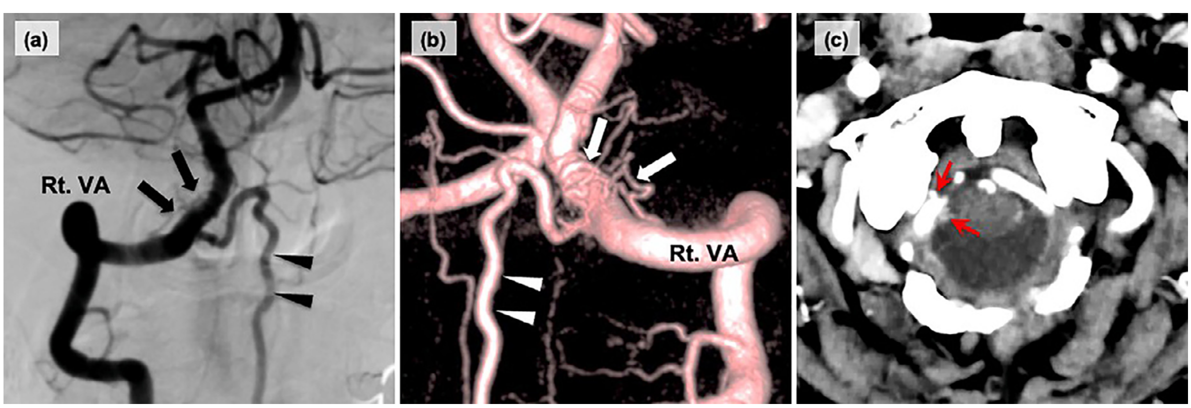

Fig. 3 – The right vertebral angiogram. The A-P view (A) and posterior view of the 3D-DSA (B) demonstrate an arteriovenous fistula at the craniocervical junction fed by right meningeal arteries (arrows) from the right VA, draining into the dilated ASV (arrowheads). The 4DCTA, axial view (C) reveals fistula points which locate around the site of the dural penetration of the right VA (red arrows). A-P, anterior-posterior; 3D-DSA, 3-dimensional digital subtraction angiography; VA, vertebral artery; ASV, anterior spinal vein; 4DCTA, 4-dimensional computed tomography angiography (Color version of figure is available online).

fore, surgical treatment was selected for the lesion. Based on the findings of the DSA, the transcondylar fossa approach for obliteration of the draining vein for CCJDAVF was attempted. Under general anesthesia, in the park-bench position, the transcondylar fossa approach were performed. The meningeal artery from the right VA and the dilated and reddish ASV were identified and exposed. Indocyanine green video angiography confirmed the feeding arteries, fistulas, and draining vein. Thereafter, the draining vein was coagulated and amputated. During the surgery, motor evoked potentials and somatosensory evoked potentials were monitored, and no abnormal findings were observed.

The postoperative course was uneventful. The patient became ambulatory three months after the surgery, although sensory disturbance in all extremities remained. Postoperative 4DCTA revealed the disappearance of the dilated ASV, and T2 weighted images showed reduction of the intramedullary high-intensity signal and disappearance of flow voids around the spinal cord. The ASL images confirmed the complete disappearance of the abnormal high-intensity signal (Fig. 2B). The patient was followed up every 6 months and no exacerbation of symptoms was observed. The ASL images did not show a high-intensity signal at the operative site.

Discussion

Diagnosis of CCJDAVF

A CCJDAVF is one of the DAVFs in which the main fistula-point is located around the craniocervical junction and that has been reported to be about 3.3% of all intracranial and spinal DAVFs. [9] Among the reported cases, it has been found to cause subarachnoid hemorrhage or progressive severe myelopathy. According to the literature, 37.1% of the patients showed myelopathy, 43.1% had subarachnoid hemorrhage, and 3.3 % had brainstem dysfunction. [10] Prompt diagnosis, appropriate treatment, and precise and careful follow-ups are mandatory.

For the neurosurgical management of DAVF, multimodal and repetitive neuroradiological methods, such as computed tomography, MRI, and DSA, are required. In considering the patient's general condition, repeatable radiological examinations without exogenous contrast medium or radiation exposure can be highly beneficial. Recently, the MRI sequence to depict CBF, ASL has been developed. The ASL is a technique to obtain information about CBF by utilizing electromagnetically labeled arterial blood as an endogenous tracer instead of an exogenous tracer such as a contrast medium. [1,2,3,4] As there is no need to use an exogenous contrast medium, ASL can be used for patients irrespective of general condition, renal function, or contrast medium allergy. The ASL is a completely noninvasive examination, which leads to enable repeated examinations. In combination with other modalities, ASL is effective in the diagnosis of brain tumors, epilepsy, and stroke has been reported. [2,3,4] It has also been described that ASL is useful for the evaluation of intracranial DAVF, such as transverse-sigmoid sinus DAVF and cavernous sinus DAVF, to predict draining veins. To confirm persistent obliteration of the fistula and absence of blood flow in the draining veins after treatment, angiography is inevitable. However, in combination with angiography and ASL, it can also be a great helpful tool for this purpose. [5,6,7,8]

An exact study protocol was not established because of the rarity of CCJDAVF and the lack of reports of using ASL in CCJDAVF diagnosis. We performed ASL imaging with two PLD settings (1.5s and 2.5s) and obtained clear images before and after treatment with the PLD = 1.5s setting but unclear with the 2.5s. This indicates that it could cause false negatives depending on the PLD settings in the evaluation for CCJDAVF with ASL. In order to evaluate this disease accurately by using ASL, it is necessary to set up multiple PLDs and study each case individually.

Neuroradiological aspects of ASL

A high-intensity signal in ASL is considered as a reflection of rapid transit time through the vasculature, also as consistent with congestion or dilated veins associated with DAVF. [5,6,7,8] Although the exact mechanism underlying the presence of a hyper-intensity signal in ALS is unknown, it is inferred to be as follows. Under normal conditions, the ASL marked high-intensity area is not seen in the typical venous system. The ASL uses electromagnetically labeled arterial blood as diffusible water, which is extracted by 90% on the first pass through the capillary bed. However, at the arteriovenous shunt in the DAVF, a capillary bed is lacking, resulting in no water extraction and shortening of the transit time of the labeled water. These factors can induce a high-intensity ASL signal in the venous system. [11,12,13] The reason for high-intensity signals in ASL in the CCJDAVF are considered to be the same as other intracranial DAVFs, and it has been suggested that the findings of ASL reflect congested ASV by AV shunt. Moreover, the disappearance of the hyper-intensity signals can be confirmed after successful interruption of the draining vein. This clearly suggests that the appearance of a hyper-intensity signal in ASL is due to the existence of an arteriovenous shunt.

Future perspective

There are a few limitations in our analysis of ASL data. First, this is the only case that shows ASL data of CCJDAVF, further data accumulation for more precise analysis is required. Second, since the craniocervical junction may be out of the FOV in routine MRI, attention must be paid to setting the FOV. Third, the PLD may cause false-negative findings in ASL. To obtain precise images in ASL, modification and devising of PLDs changes may be required. In addition, it is necessary to evaluate ASL images with other modalities such as DSA and/or 4DCTA. [7]

Conclusion

The ASL is a useful noninvasive tool for depicting a high blood-flow intracranial lesion, such as DAVF. CCJDAVF, high-flow shunts, and dilated draining veins are well-identified on ASL. To obtain precise ASL images of CCJDAVF, some modifications and devising in MRI studies are necessary. The noninvasive nature of ASL can be beneficial to patients with CCJDAVF for long-term post-treatment follow-up.

Patient consent

Informed consent was obtained from the patient about the publication of the case details and any associated images.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2021.04.006.

REFERENCES

[1] Williams DS, Detre JA, Leigh JS, Koretsky AP, Koretsky AP. Magnetic resonance imaging of perfusion using spin inversion of arterial water. Proc Natl Acad Sci USA 1992;89:212–16.

[2] Deibler AR, Pollock JM, Kraft RA, Tan H, Burdette JH, Maldjian JA. Arterial spin-labeling in routine clinical practice, part 1: technique and artifacts. AJNR Am J Neuroradiol 2008;29:1228–34.

[3] Deibler AR, Pollock JM, Kraft RA, Tan H, Burdette JH, Maldjian JA. Arterial spin-labeling in routine clinical practice, part 2: hypoperfusion patterns. AJNR Am J Neuroradiol 2008;29:235–1241.

[4] Deibler AR, Pollock JM, Kraft RA, Tan H, Burdette JH, Maldjian JA. Arterial spin-labeling in routine clinical practice, part 3: hyperperfusion patterns. AJNR Am J Neuroradiol 2008;29:1428–35.

[5] Noguchi K, Kawabe H, Seto H. Intracranial dural arteriovenous fistula: preliminary report of arterial spin labeling. Open J Med Imaging 2013;3:1–6.

[6] Sakamoto S, Kiura Y, Okazaki T, Shinagawa K, Miyoshi H, Kurisu K. Arterial spin-labeling imaging at 3-T in dural arteriovenous fistulas of cavernous sinus before and after endovascular treatment. Hiroshima J Med Sci 2012;61:105–107.

[7] Yamamoto N, Yamamoto Y. Dural arteriovenous fisutula at the cavernous sinus diagnosed by arterial spin-labeled imaging. Intern Med 2018;57:1163–6.

[8] Le TT, Fischbein NJ, Andre JB, Wijman C, Rosenberg J, Zaharchuk G. Identification of venous signal on arterial spin labeling improves diagnosis of dural arteriovenous fistulas and small arteriovenous malformations. AJNR Am J Neuroradiol 2012;33:61–8.

[9] Satomi J, Satoh K. Epidemiology and etiology of dural arteriovenous fistula. Brain Nerve 2008;60:883–6.

[10] Wang JY, Molenda J, Bydon A, Colby GP, Coon AL, Tamargo RJ, et al. Natural history and treatment of craniocervical junction dural arteriovenous fistulas. J Clin Neurosci 2015;22:1701–7.

[11] Wolf RL, Wang J, Detre JA, Zager EL, Hurst RW. Arteriovenous shunt visualization in arteriovenous malformations with arterial spin-labeling MRI. AJNR Am J Neuroradiol 2008;29:681–7.

[12] Kukuk GM, Hadizadeh DR, Bostrom A, Gieseke J, Bergener J, Nelles M, et al. Cerebral arteriovenous malformations at 3.0 T: intraindividual comparative study of 4D-MRA in combination with selective arterial spin labeling and digital subtraction angiography. Invest Radiol 2010;45:126–32.

[13] Pollock JM, Whitlow CT, Simonds J, Stevens EA, Kraft RA, Burdette JH, et al. Response of arteriovenous malformations to gamma knife therapy evaluated with pulsed arterial spin-labeling MRI perfusion. AJR Am J Roentgenol 2010;196:15–22.