

## Case Report

## Usefulness of preoperative cone beam computed tomography and intraoperative digital subtraction angiography for dural arteriovenous fistula at craniocervical junction: Technical case report

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### Abstract

**Background:** Direct surgery is commonly selected for the treatment of cranio-cervical junction dural arteriovenous fistula and its outcome is more satisfactory than that of embolization. Intraoperative treatment evaluation is relatively easy in embolization, whereas in direct surgery it can be difficult.

**Case Description:** A 67-year-old male suffered a subarachnoid hemorrhage. On three-dimensional (3D) images of preoperational cone-beam computed tomography (CBCT), the structure of the draining vein was depicted in detail along with the surrounding bone structures. The radial artery penetrated the dura mater, and it was found that there were two veins derived from the radiculospinal vein; one was the anterior radicular vein descending toward the dorsal side (the shallow layer of the surgical field) and the other was the anterior spinal medullary vein ascending toward the ventral side (the deep layer of the surgical field) and flowing out to the anterior spinal vein.

**Conclusion:** Without detailed assessments with preoperative CBCT, the surgery might have been done with dissection of only the anterior radicular vein in the shallow layers. For identification of the draining vein located deep in the surgical field, such as the cranio-cervical junction, careful assessments using 3D CBCT images are important.

**Key Words:** Cranio-cervical junction, direct surgery, dural arteriovenous fistula, intraoperative digital subtraction angiography, preoperative cone beam computed tomography

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## INTRODUCTION

Direct surgery is commonly selected for the treatment of cranio-cervical junction dural arteriovenous fistula (CCJ-dAVF) and its outcome is more satisfactory than that of embolization.<sup>[49,52]</sup> Intraoperative treatment evaluation is possible in embolization, whereas in the direct surgery it is often performed using postoperative digital subtraction angiography (DSA). In this study, we experienced a case where preoperative cone beam computed tomography (CBCT) and DSA during direct surgery were useful in treatment; hence, we report this case along with a literature review.

## CASE REPORT

The patient was a 67-year-old male who developed the condition with sudden posterior cervical pain and vomiting before going to bed. As headache and nausea were still present on the following day, he visited the general outpatient clinic by walking. During the physical examination, the patient was clearly conscious without any neurologic deficit symptoms or meningeal irritation symptoms. On head CT, subarachnoid hemorrhage (SAH) was observed [Figure 1]. On the subsequent DSA of the left vertebral artery, a slightly abnormal shadow from the meningeal branch was detected, however, it was not concluded as the source of bleeding. The patient received conservative treatment, and on day 15 of illness, DSA was repeated. On angiography of the left vertebral artery, the patient was diagnosed with dAVF, with which blood flew in from the radicular artery of the left vertebral artery and flew out to the anterior spinal vein at the level superior to the C1/Foramen magnum [Figure 2a]. CBCT was performed using an Allura Xper FD20/10<sup>®</sup> angiography device (Philips Electronics Japan). The images were obtained at a tube voltage at 80 kv, 30 fr/s using nondiluted contrast medium at 1.0 ml/s for 25 s. On Xtra



**Figure 1:** Computed tomography (CT) shows subarachnoid hemorrhage (SAH)

Vision workstation, Maximum Intensity Projection (MIP) images were reassembled to obtain detailed depiction of the structure of the draining vein along with the surrounding bone structures in three-dimensional (3D) vision. The radial artery was bifurcated to the one flowing out to the epidural venous plexus before penetrating the dura mater and to the one penetrating the dura mater. Once it penetrated the dura mater, it was found that there were two veins derived from the radiculospinal vein; one was the anterior radicular vein descending toward the dorsal side (the shallow layer of the surgical field) and another was the anterior spinal medullary vein ascending toward the ventral side (the deep layer of the surgical field) and flowing out to the anterior spinal vein at the level of foramen magnum while forming the coronal venous plexus [Figure 2b-e]. On day 23 of illness, dissection of the draining vein was performed with craniotomy.

## Surgical procedures / intraoperative findings

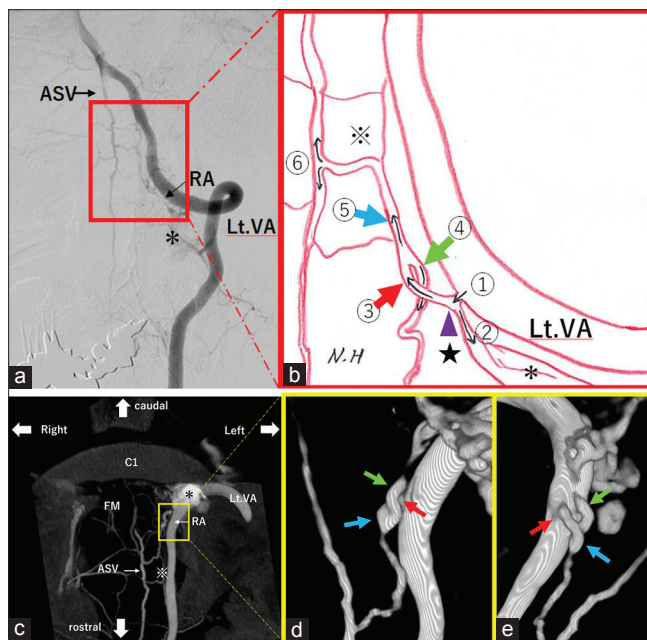
As intraoperative DSA (C-arm) had been scheduled at the same time, a 30 cm-long 4-Fr sheath was placed in the left femoral artery in the supine position under general anesthesia. The head was fixed using MAYFIELD<sup>®</sup>, which is made of carbon so that it does not affect DSA. Subsequently, lateral suboccipital craniotomy and ipsilateral C1 laminectomy were performed in the prone position. The C1 nerve root was observed near the left vertebral artery, and after dissecting the dura mater, the spinal branch of the accessory nerve and C1 nerve root were observed inside the dura mater. The anterior radicular vein was observed dorsal to the site where the vertebral artery penetrated the dura mater [Figure 3a]. A catheter was placed in the left vertebral artery, and intra-arterial injection of indocyanine green (ICG) was performed. It was confirmed that the anterior radicular vein was depicted in the arterial phase [Figure 3b]. This was temporarily blocked using a temporary clip. However, the draining vein was still observed on the intraoperative DSA. Furthermore, the radiculospinal vein bifurcating to the anterior spinal medullary vein in the deep surgical field was identified [Figure 3c]. This was also temporarily blocked using a temporary clip, and it was confirmed that the draining vein was not observed on the intraoperative DSA performed for the second time [Figure 3d]. After cauterizing the anterior radicular vein and the radiculospinal vein, DSA was repeated to ensure that dAVF had been dissected. Then, the wound was closed. The patient was placed in the supine position again and the 4Fr sheath was removed. Manual compression was performed at the completion of the surgery.

## Postoperative course

No neurological deficit symptom was observed after the operation. On angiography on day 34 of illness, dAVF had been disappeared, and on day 37 the patient was discharged; he went back home by walking.

## DISCUSSION

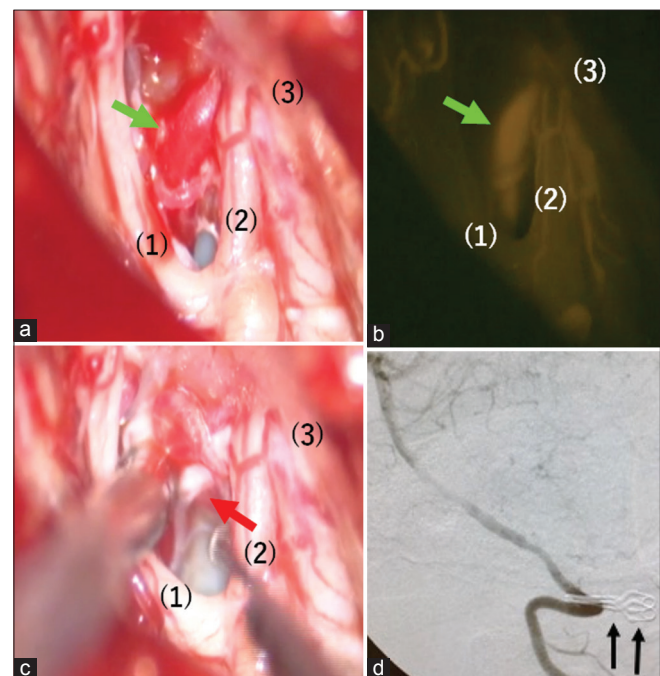
CCJ-dAVF does not have a venous sinus, which should be embolized, and its feeding artery, which is usually the vertebral artery, is close to the site of the fistula. Therefore, it is not easy to perform intravascular embolization.<sup>[30,40,43]</sup> There are also some reports regarding the outcomes of spinal dAVF; the occlusion rates were 98% for direct surgery and 46% for intravascular embolization in one report<sup>[38]</sup> and 96.6% for direct surgery and 72.2% for intravascular embolization in another.<sup>[4]</sup> Direct surgery is the only independent prognostic factor<sup>[49]</sup> of CCJ- dAVF for a satisfactory outcome. However, it is not easy to obtain accurate intraoperative treatment evaluation using direct surgery without assistance. In a single surgery without assistance, it was found sometimes on the postoperative DSA that the draining vein, which was supposed to be dissected, was actually not dissected. Hence, it is desirable to use surgical assistance.<sup>[30]</sup> Diagnostic imaging by preoperative DSA alone may not be adequate to accurately understand the circulatory dynamics.<sup>[27]</sup>



**Figure 2:** (a): Digital subtraction angiography (DSA) (Anterior-Posterior view) reveals arteriovenous fistula at craniocervical junction. VA: Vertebral Artery, RA: Radicular Artery, ASV: Anterior Spinal Vein (b): Schematic illustration shows detail flows feeding artery to draining vein. (c): Cone beam CT (CBCT) demonstrates the vascular anatomy and clearer definition of the relationships to bone structure. (d and e): Radiculospinal vein is on the back side of anterior radicular vein (d: posterior-anterior view, e: 180 degrees rotated Figure 2d) (\*) Epidural Venous Plexus (EVP), ① Radicular artery, ② Radicular artery flows into EVP, (★) Fistulous site (purple arrowhead), ③ Radiculospinal vein (red arrow), ④ Anterior radicular vein (green arrow), ⑤ Anterior spinal medullary vein (blue arrow), (※) Coronal Venous Plexus (CVP), ⑥ Anterior spinal vein, FM: Foramen Magnum, VA: Vertebral Artery, RA: Radicular Artery, ASV: Anterior Spinal Vein

Accurate preoperative imaging evaluation is essential for surgical planning. Vaso CT is very useful for preoperative imaging evaluations, and there are some reports of the use of Dyna CT<sup>[20]</sup> and CBCT<sup>[14]</sup> for cranial dAVF and CBCT for spinal dAVF.<sup>[1]</sup> Dyna CT and CBCT can reveal hemodynamic characteristics of dAVF and anatomical details of fistulas, and can clearly depict the relationship with the surrounding bone structures.<sup>[1,20]</sup> These enable us to obtain 3D anatomical structures and easily visualize each anatomical position through reconstruction with arbitrary sections/width of slices. High-resolution images can be obtained using a low-dose contrast medium.<sup>[46]</sup>

CBCT can even depict AVM nidus behind a hematoma.<sup>[35]</sup> It is useful not only in diagnosis of micro AVMs but also in posttreatment follow up.<sup>[7]</sup> In our case, detailed 3D assessment of preoperative CBCT revealed that there were two draining routes, suggesting that the radiculospinal vein in the deep layer of the surgical field might be located behind the anterior radicular vein in the shallow layer of the surgical field. There are also some reports of intraoperative use of Dyna CT<sup>[23]</sup> and 3D rotational angiography (3DRA).<sup>[12]</sup> As these were not available at our facility, we decided to perform intraoperative DSA. It has been also reported that early venous filling, sinus occlusion, leptomeningeal venous drainage, and varices can be detected using magnetic



**Figure 3:** Intraoperative view. (a): Anterior radicular vein exhibits red vein (green arrow). (b): Anterior radicular vein exhibits arterial flow by injecting ICG (green arrow). (c): Radiculospinal vein is on the back side of anterior radicular vein and collapsing by suction tube (red arrow). (d): Digital subtraction angiography (DSA) (lateral view) reveals disappearance of venous drainage route and two temporary clips are reflected (black arrows). (1) spinal cord, (2) C1 nerve root, (3) dura matter

resonance digital subtraction angiography (MRDSA), which identifies hemodynamic abnormalities associated with bleeding risks.<sup>[21]</sup> Meanwhile, for CT and MRI, it has been reported that there is a limit in the classification and diagnosis of dAVF, and that it is not possible to obtain detailed anatomical depictions for all structures.<sup>[20]</sup> 3D time-of-flight MRA can depict abnormal arterial flow and venous abnormalities but can hardly identify the site of the fistula.<sup>[28]</sup> Selective DSA requires a large amount of contrast medium, resulting in longer examination time and greater level of radiation exposure.<sup>[1,20]</sup> These problems with DSA can be minimized with Dyna CT and CBCT. Although 3DRA is essential for detailed vascular information, depiction of the surrounding bone structures is ambiguous compared to CT angiography (CTA).<sup>[1,8,19,20]</sup> There are also reports of 3D computer graphics (3DCG) which combine 3DRA and postmyelographic CT<sup>[41]</sup> as well as a report that MPR images of 3DRA were effective for identification of fistula sites.<sup>[47]</sup>

Surgical assistance for vascular lesions include color flow Doppler, ICG, DSA, navigation,<sup>[2]</sup> and arterial injection of selective pigments.<sup>[42,45]</sup>

Doppler is an easy and minimally invasive method which is commonly used for observation of blood flow, but it may not detect the pulse wave accurately due to pulsation, pooling of cerebrospinal fluid/blood, or the angle of the probe. Moreover, it can evaluate only up to cortical arteries; blood flow of small perforating branch cannot be evaluated.<sup>[3]</sup> In cerebral aneurysm surgery, it is difficult to evaluate very small blood-flow residue inside the aneurysm, and it is not possible to detect stenosis which does not affect Doppler signals.<sup>[16,39]</sup>

ICG videoangiography is also an easy and minimally invasive surgical assistance which is useful in both cranial/spinal dAVF.<sup>[36]</sup> The dose of intravenous ICG injection is 0.2–0.5 mg/kg and limited up to 5 mg/kg/day.<sup>[22,24,33]</sup> The incidence of adverse events is 0.2%<sup>[22]</sup> and requires 10–20 minutes of interval until reexamination.<sup>[22,24,36]</sup> There is a concern that the dose is likely to increase in a shunt disease, which may require repetitive intravenous administrations. While with intra-arterial administration, the dose can be small, and the lesion can be depicted in a shorter period of time, and more precisely, there is an advantage that the dose will never be excessive even with repetitive administrations.<sup>[22,50]</sup> A real-time blood flow evaluation is possible with ICG during a surgery under direct observation. As long as it is within the range of visual axis movement, blood flow can be evaluated from multiple directions with one dose of ICG. As it clearly depicts the perforating branch, it can function as a probe facilitating safe dissection.<sup>[26]</sup> Evaluations can be made by a large number of people on a video monitor. It is, however, difficult to observe under the presence of hematoma and to make accurate evaluations if the

thickness of wall is inconsistent due to calcification of the vascular walls. Moreover, areas that are hidden in the surgical field cannot be evaluated. In cases with deep lesions out of 31 cases with cerebral arteriovenous malformation (AVM), two lesions, which were difficult to be depicted using ICG alone, were newly detected by adding intraoperative DSA.<sup>[6]</sup> Attention is needed as deep thalamic AVM was not detected with ICG alone.<sup>[17]</sup>

Considering the limit of ICG for deep thalamic AVM,<sup>[15,31,51]</sup> addition of intraoperative ultrasound scan has been also suggested.<sup>[48]</sup> In cerebral aneurysm surgery, it is difficult to observe the bifurcation of internal carotid artery and ophthalmic artery with ICG,<sup>[37]</sup> resulting in inadequate evaluation of the deep lesion.<sup>[11]</sup> It has been also reported that neuroendoscopic observation can be useful for areas hidden under the microscopic field.<sup>[29]</sup>

It is advantageous for intraoperative DSA that it can evaluate not only the surgical field but also extensive cerebral vascular conditions with a large number of people. However, it requires arterial puncture and complex operation of the imaging device. Moreover, it takes time to conduct procedures, resulting in prolongation of the operation time;<sup>[5]</sup> the costs for the medical devices and facilities are also high.<sup>[18]</sup> There is also a concern regarding radiation exposure; deterioration of renal functions may occur due to use of contrast medium multiple times in contrast radiography from various directions. As it is performed during the operation, the direction of projection may be restricted; it is likely to be affected by metal devices in the surgical field as well. The perforating branch cannot be observed, and permanent neurological sequela may occur at 0.35–1.5% due to thromboembolism.<sup>[10,13,44]</sup>

Out of patients who received routine DSA during cerebral aneurysm surgery, only 20% required intraoperative DSA.<sup>[25]</sup> Unlike Doppler and ICG, DSA cannot evaluate blood vessels under direct observation; hence, it is considered inferior to ICG in terms of identification of blood vessels in the surgical field.<sup>[24]</sup> In cerebral aneurysm surgery, even if there is no abnormality detected in intraoperative DSA, aneurysm neck remnant was observed in 7.9% and bifurcation vascular occlusion in 4.8% in postoperative DSA.<sup>[32]</sup>

In our patient, a catheter was placed in the left vertebral artery under X-ray immediately before intraoperative DSA. Although it is easier to place the catheter in the supine position right after induction of general anesthesia, we performed catheterization in the prone position immediately before taking the images due to concerns about thromboembolism induced by long-term catheter placement. It might have been easier to perform the left brachial artery puncture rather than the left femoral artery puncture. One study reported that CCJ-dAVF was

cured using ICG alone.<sup>[5]</sup> For the use of ICG in direct surgery for dAVF, it has been reported that intraoperative DSA was performed concomitantly in all patients of 13 cranial dAVF cases, and for 12 cases of spinal dAVF, DSA or MRI/MRA was performed postoperatively in all patients. All patients were cured, and for some patients with cranial dAVF, the results of vascular evaluations were similar for intraoperative ICG and intraoperative DSA.<sup>[36]</sup> There are advantages and disadvantages for intraoperative Doppler, ICG, and DSA;<sup>[34]</sup> there are reports stating that ICG and Doppler should be used concomitantly to observe complementary<sup>[16]</sup> and that ICG should be used in combination with DSA or Doppler.<sup>[9,11]</sup> In our patient, the anterior radicular vein was depicted in the arterial phase with intraarterial injection of ICG from the left radial artery, whereas the radiculospinal vein, which was located even deeper was not detected with ICG. It was assumed that similar results would have been obtained even if intraarterial injection of indigo carmine<sup>[45]</sup> was performed from the left vertebral artery. The draining vein is macroscopically a red vein and is visually recognizable in general. However, if blood vessels are overlapped, as in this case, visual recognition of the red vein may be difficult as the blood vessel in the deep layer is compressed when the blood vessel in the superficial layer is compressed to visualize the deep vein, suggesting that the effectiveness of ICG or indigo carmine would be poor. Namely, preoperative understanding of 3D structures would have been necessary.

## CONCLUSION

We experienced a case which received direct surgery for cranio-cervical junction dural arteriovenous fistula induced by subarachnoid hemorrhage. Deliberate assessment of 3D structures of CBCT was crucial for identification of the draining vein if the surgical field is located deep such as in the cranio-cervical junction.

## Declaration of patient consent

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

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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