



Non-Sinus-Type Dural Arteriovenous Fistula at the Foramen Magnum: A Review of the Literature

Masafumi Hiramatsu,¹ Tomohiko Ozaki,² Rie Aoki,³ Shinri Oda,³ Jun Haruma,¹ Tomohito Hishikawa,¹ Kenji Sugiu¹ and Isao Date¹

Dural arteriovenous fistula (dAVF) of the foramen magnum (FM) region is rare. Moreover, the terminology of dAVF is very confusing in this region. In the narrow sense, the FM dAVF is the non-sinus-type dAVF with direct venous reflux to the medulla oblongata or spinal cord via the bridging veins (BVs) of the FM. Previous literature was systematically reviewed to investigate the clinical characteristics, angioarchitecture, and effective treatment of the FM dAVF. From the literature review, almost all the feeders of FM dAVF were dural branches. Spinal pial arteries were rarely involved as the feeder. All lesions had venous reflux to the medulla oblongata via medullary BVs. The FM dAVF is characterized by a significant male predominance and a high incidence of aggressive symptoms. The most common symptom is congestive myelopathy, followed by hemorrhage. The FM dAVF differs from the craniocervical junction (CCJ) arteriovenous fistula (AVF) and is similar to the thoracolumbar spinal dAVF. Direct surgery for the FM dAVF is effective and safe. Endovascular treatment for the FM dAVF may be more effective and has lower complication rates than that for the CCJ AVF.

Keywords ▶ dural arteriovenous fistula, foramen magnum, bridging vein, treatment

Introduction

Dural arteriovenous fistulas (dAVFs) are relatively rare diseases, and the recently reported crude incidence rate was 1.044 per 100000 person-years.¹⁾ In the cranial dAVF, the foramen magnum (FM) dAVF is an extremely rare disease, with few reports on its characteristics and effective treatment. In the present review article, we attempted to define the FM dAVF in a narrow sense. Moreover, we investigated the characteristics, angioarchitecture, and effective treatment of FM dAVF using a systematic literature review.

¹Department of Neurological Surgery, Okayama University Faculty of Medicine, Dentistry and Pharmaceutical Sciences, Okayama, Japan

²Department of Neurosurgery, National Hospital Organization Osaka National Hospital, Osaka, Japan

³Department of Neurosurgery, Tokai University Hachioji Hospital, Tokyo, Japan

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Corresponding author: Masafumi Hiramatsu. Department of Neurological Surgery, Okayama University Faculty of Medicine, Dentistry and Pharmaceutical Sciences, 2-5-1, Shikatacho, Kita-ku, Okayama, Okayama 700-8558, Japan
Email: mhiramatsu@okayama-u.ac.jp



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dAVF around the FM region and confusion about the terminology

The terminology of dAVF in this region is confusing, with various dAVFs being referred to in various ways. Li et al. have written a review article regarding the dAVFs of the lateral FM region.²⁾ They divided the lateral FM dAVFs into four types: anterior condylar confluence dAVF, posterior condylar canal dAVF, marginal sinus (MS) dAVF, and jugular foramen dAVF. McDougal et al. reported dAVFs of the MS and divided them into three grades as follows: grade 1, with unrestricted drainage; grade 2, with restricted drainage; and grade 3, with retrograde cortical drainage.³⁾ Caton et al. reported dAVFs of the FM region and divided them into four groups as follows: type 1, with unrestricted sinus drainage; type 2, with sinus reflux; type 3, with reflux involving sinuses and cortical veins; and type 4, with restricted cortical vein outflow or perimedullary congestion.^{4,5)} Spittau et al. reported a systematic literature review of similar cases as the dAVFs of the hypoglossal canal and divided them into three groups as follows: type 1, with anterograde drainage; type 2, with retrograde drainage to the cavernous sinus; and type 3, with cortical and/or perimedullary drainage.⁶⁾ In short, the above authors analyzed similar lesions, including anterior condylar arteriovenous fistulas (AVFs), and divided them using the classification proposed by Borden et al.⁷⁾ Recently, it has been found that

most anterior condylar AVFs have intraosseous shunts that drain to the surrounding veins via the anterior condylar vein (ACV)⁸⁾ and is considered a different disease than the FM dAVF in the narrow sense. Similar to the anterior condylar AVF, most posterior condylar canal dAVFs have intraosseous shunts that drain to the surrounding veins via the posterior condylar vein in the posterior condylar canal.⁹⁾

So, what kind of lesions are FM dAVFs in the narrow sense? The FM has a rich surrounding venous network, which includes the MS, ACV, suboccipital cavernous sinus (SCS), and occipital sinus. These veins or sinuses have various connections to the veins of the medulla oblongata via the bridging veins (BVs).¹⁰⁾ Mitsuhashi et al. reported the dAVFs draining into the BV of the medulla.¹¹⁾ They included three cases of dAVFs at the occiput–C1 level and two cases of dAVFs at the FM. All cases had medullary or spinal venous drainage, and most patients presented with subarachnoid hemorrhage (SAH) or venous hypertension of the spinal cord. Motebejane et al. reported the 12 cases series of the FM dAVF.¹²⁾ They described that all the dAVFs were located in the lateral epidural space on the margin of the FM, and most cases presented with paraparesis or SAH. Recently, Yoo et al. reported medullary BV-draining dAVFs and divided them into nine FM and 13 craniocervical junction (CCJ) lesions.¹³⁾ All cases had medullary or spinal venous drainage, and most presented with hemorrhage or myelopathy. In summary, the FM dAVF, in the narrow sense, is the non-sinus-type dAVF with direct venous reflux to the medulla oblongata or spinal cord via the BVs of the FM. Because the non-sinus-type FM dAVF has venous reflux to the medulla oblongata, it can be more aggressive than the sinus-type dAVF, such as anterior condylar AVF. Moreover, transvenous embolization, the standard procedure for sinus-type dAVF, may be difficult to treat non-sinus-type FM dAVF.

Literature review of the non-sinus-type FM dAVF

Previous literature was systematically reviewed to investigate the clinical characteristics, angioarchitecture, and effective treatment of FM dAVF. Using the PubMed database, we searched case reports and case series written in English and published between 2000 and 2022 with the term (((“foramen magnum”) AND (arteriovenous)) AND (“2000”[Date - Publication]: “2022”[Date - Publication])) AND (English [Language]). Cases of dAVFs located at the FM were included in this analysis. The anterior condylar, posterior condylar canal AVFs, or CCJ AVFs at the C1 or lower spine level were excluded. Moreover, although there can be some FM dAVFs in the case reports regarding the

Table 1 Summary of the literature review regarding the FM dural arteriovenous fistula

Characteristics	Value
Total number of patients	56
Age (years)	
Median	54
Mean	54
Male	52 (93%)
Presentation	
Myelopathy	29 (52%)
Hemorrhage	21 (38%)
SAH	16 (29%)
ICH	3 (5%)
IVH	2 (4%)
Feeding artery	
VA	38 (68%)
APA	37 (66%)
OA	15 (27%)
ASA	0 (0%)
LSA	1 (2%)
Aneurysm	0 (0%)
Draining vein	
Reflux to the medullary vein	56 (100%)
Ascending venous drainage	15 (27%)
Descending venous drainage	27 (48%)
Both venous drainage	12 (21%)
Varix	22 (39%)
Treatment	
Embolization only	37 (66%)
DS after embolization	5 (9%)
DS only	13 (23%)
Complication	
After EVT	3/42 (7%)
After DS	0/13 (0%)

APA: ascending pharyngeal artery; ASA: anterior spinal artery; DS: direct surgery; EVT: endovascular treatment; ICH: intracerebral hemorrhage; IVH: intraventricular hemorrhage; LSA: lateral spinal artery; OA: occipital artery; SAH: subarachnoid hemorrhage; VA: vertebral artery

CCJ AVF, we did not include these cases in the literature review.

The literature review identified 56 FM dAVFs from six case series^{11–16)} and 21 case reports (**Table 1**).^{17–37)} The median and mean ages were 54 and 54 years, respectively. There were 52 males (93%) and four females (7%). Twenty-nine patients (52%) presented with congestive myelopathy, and 21 (38%) presented with hemorrhage. In the hemorrhagic cases, 16 patients (29%) presented with SAH, three patients (5%) presented with intracerebral hemorrhage (ICH), and two patients (4%) presented with intraventricular hemorrhage. As the ICH in three patients, hemorrhagic locations were the cerebellum, occipital lobe, and medulla oblongata. One patient presented with a neurological deficit due to the mass effect on the medulla oblongata by venous varix. Two patients presented with other neurological symptoms,

such as trigeminal neuralgia and gaze paresis. Non-specific symptoms, such as headache, appeared in three patients. Onset-to-diagnosis duration was identified in 33 patients. In the patients with myelopathy, the median duration was 7 months, and the range was 14 days to 10 years. In the patients with hemorrhagic presentation, the median duration was 2 days, and the range was 0 to 13 days.

As the feeding artery, the radicular or meningeal arteries from the vertebral artery (VA) were involved in 38 lesions (68%). No lesions were supplied by the anterior spinal artery (ASA), and one lesion was supplied by the lateral spinal artery (LSA). One lesion was supplied by the posterior inferior cerebellar artery. The ascending pharyngeal artery (APA), occipital artery (OA), and posterior auricular artery were involved in 37 (66%), 15 (27%), and one lesion, respectively. There was no aneurysm on the feeder.

As the draining vein, all lesions had reflux venous drainage to the medulla oblongata. Fifteen lesions (27%) had ascending venous drainage via medullary veins, 27 lesions (48%) had descending venous drainage to the spinal veins, and 12 lesions (21%) had venous drainage in both directions. Two lesions had venous drainage to the medulla oblongata, and we could not identify the ascending or descending drainage routes. There were 22 lesions (39%) with venous varix.

As the first-line treatment, endovascular treatment (EVT) and direct surgery (DS) were selected in 42 (75%) and 13 (23%) lesions. All EVTs were transarterial embolization (TAE) using liquid materials, such as n-butyl-2-cyanoacrylate (NBCA), Onyx, PHIL, or Squid. Twenty-three lesions were treated by NBCA, and 14 lesions were treated by Onyx. Thirty-seven lesions (66%) were obliterated by EVT only, and five (9%) treated with EVT required DS to obliterate. All lesions treated with the DS were obliterated. One lesion was not treated.

The postoperative course was described in 42 patients. After treatment, there were 31 patients (74%) with good recovery, including modified Rankin scale of 0 to 1. Four patients (10%) had moderate or severe disabilities during the follow-up period. Six patients were described as improved. One patient presented with myelopathy was defined as having no change.

Angioarchitecture of the non-sinus-type FM dAVF

In **Fig. 1**, we show the representative case of the FM dAVF. From the literature review, almost all feeders were dural branches from VA, APA, and OA. There are some dural branches from VA, such as the radiculomeningeal artery, anterior meningeal artery, and posterior meningeal artery.

The APA has a neuromeningeal branch branching into the jugular and hypoglossal branches. The proximal portion of the OA has jugular and stylomastoid branches. These dural branches may supply the dura mater covering the FM. Unlike the CCJ AVF, involvement of the spinal arteries, such as ASA or LSA, is infrequent in the FM dAVF. In the CCJ AVF, 63% of lesions were fed by a spinal pial artery from the ASA and/or the LSA, and 32% of lesions had aneurysmal dilatation.³⁸⁾

All lesions had venous reflux to the medulla oblongata. However, it was challenging to identify the drainage route from the shunt point on the FM dura to the medulla oblongata in the past literature. Most of the drainage route could be the BV of the FM. Matsushima et al. reported the BVs from the medullary veins to the MS around the FM using a cadaveric study.³⁹⁾ They also reported the vagal vein running along the vagal nerve. Moreover, Duvernoy reported the satellite veins of the cranial nerves, such as the glossopharyngeal nerve and hypoglossal nerve.⁴⁰⁾ He also reported the satellite vein of the C-1 nerve root. These veins are the BVs connecting the medullary veins to the surrounding venous structure, such as the MS, jugular bulb, sigmoid sinus, and SCS. Based on the anatomical location, the BV of the SCS running along the C-1 nerve root can be the draining vein of the CCJ AVF. The CCJ AVF may develop along the C-1 or C-2 nerve roots and can be located on the spinal cord, spinal nerves, and/or the inner or outer surface of the dura matter.³⁸⁾ On the other hand, the BV of the MS can be the draining vein of the non-sinus-type FM dAVF. This BV does not run along the cranial nerves or spinal nerve roots. Other BVs connecting to the ACV or occipital sinus can be the draining vein of the non-sinus-type FM dAVF. Because the spinal pial arteries, such as ASA and LSA, can supply the nerve root, the CCJ AVF may be supplied by the spinal pial arteries, and the FM dAVF may not. We show the schematic illustrations of normal anatomy in **Fig. 2A** and vessel anatomy of the AVF in **Fig. 2B** with special reference to the difference between the FM dAVF and CCJ AVF.

Clinical characteristics of the non-sinus-type FM dAVF

From the literature review, the FM dAVF is characterized by a significant male predominance (93%) and a high incidence of aggressive symptoms. The most common symptom was congestive myelopathy (52%), followed by hemorrhage (38%), which together accounted for 90% of patients with aggressive symptoms. The most common hemorrhagic presentation was the SAH (16/21: 76%). These

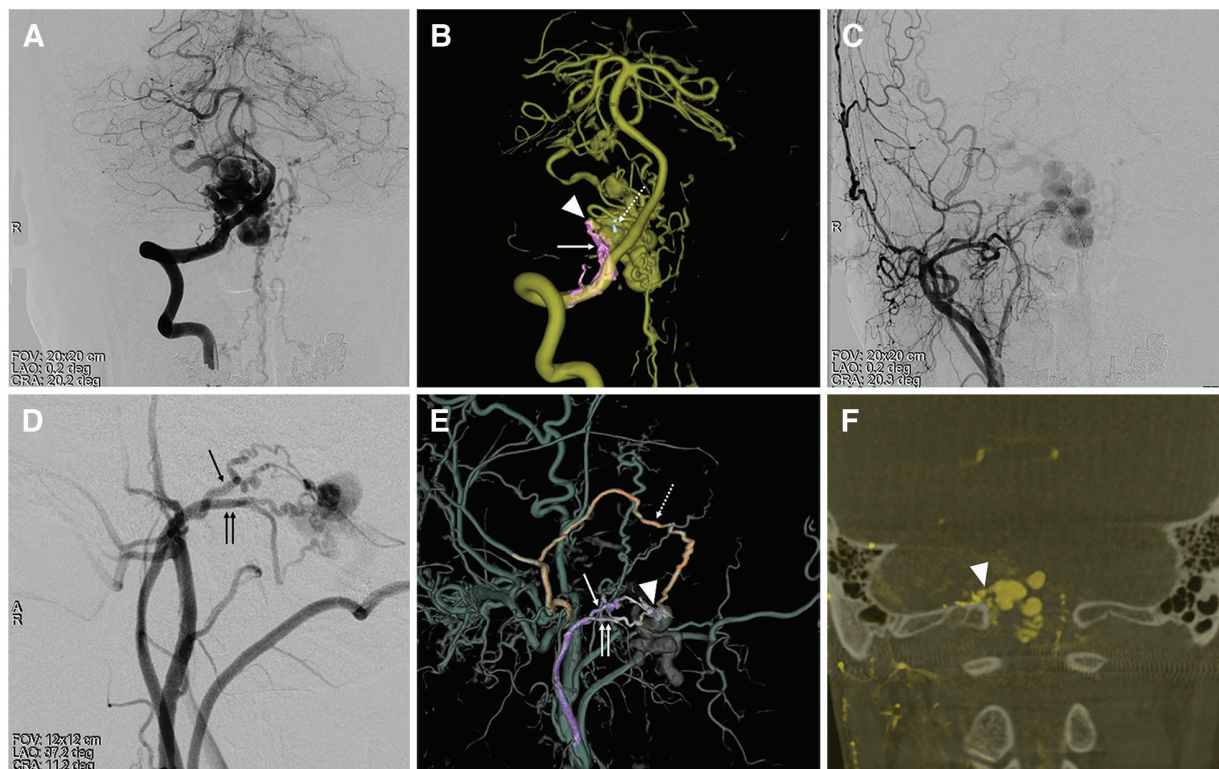


Fig. 1 A representative case of the FM dAVF. The anterior–posterior view of the 2D-DSA (A) and 3D-DSA (B) of the right vertebral angiogram shows the dAVF, which is fed by the dural branches (arrow) from the V3 segment of the VA and PICA (dotted arrow). The dAVF drains into the medullary veins with venous varices and has ascending and descending drainage routes. The arrowhead shows a shunt point. The anterior–posterior view (C) and lateral view (D and E) of the 2D-DSA (C and D) and 3D-DSA (E) of the right external carotid angiogram show the dAVF, which was fed by the jugular (arrow) and hypoglossal (double arrow) branches of the APA and the petrosquamous branch of the middle meningeal artery (dotted arrow). The coronal view of the slab maximum intensity projection image (F) from 3D-DSA of the right external carotid angiogram shows the shunt point (arrowhead) located on the upper margin of the FM. APA: ascending pharyngeal artery; dAVF: dural arteriovenous fistula; DSA: digital subtraction angiography; FM: foramen magnum; PICA: posterior inferior cerebellar artery; VA: vertebral artery

characteristics are similar to the CCJ AVF, which have male predominance (76%) and a higher rate of aggressive symptom (85%).³⁸⁾ Based on the Japanese Registry of NeuroEndovascular Therapy 2 (JR-NET2), the most male-dominated intracranial dAVF was the ACF, with 84% males.⁴¹⁾ The location of the intracranial dAVF with the highest proportion of cases occurring with aggressive symptoms was superior sagittal sinus (SSS), with an aggressive symptom rate of 57%. The male predominance and the high number of aggressive symptoms are the characteristics of the specific group of dAVF. Geibprasert et al. reported a new classification of dAVF, and the lateral epidural group includes the MS (lateral portion – FM) dAVF with the emissary-BVs of the brainstem.⁴²⁾ This lateral epidural group had male predominance (81%) and a higher rate of aggressive symptoms (86%). Tanaka reported the classification of dAVF based on embryological consideration, and the ventral and lateral parts of the MS are included in the ventral group located on the endochondral bone, and the dorsal part of the MS is included in the dorsal

group located on the membranous bone.⁴³⁾ However, the other falco-tentorial group had male predominance (87%) and a higher rate of aggressive symptoms (80%). We considered the FM dAVF to be more natural to be included in the falco-tentorial group.

Based on the literature review, 29 patients with FM dAVF presented with myelopathy; all but one patient (97%) had a descending drainage route to the spinal vein, and four patients (14%) had varix. On the other hand, 21 patients with FM dAVF presented with hemorrhage; 16 patients (76%) had ascending venous drainage via the medullary veins and 14 patients (67%) had varix. Risk factors for the hemorrhagic presentation in the CCJ AVF were reported as the involvement of ASA and the presence of aneurysmal dilatation,³⁸⁾ whereas for FM dAVF, it may be ascending venous drainage and the presence of varix.

Treatment of the non-sinus-type FM dAVF

From the literature review, 37 (66%), 5 (9%), and 13 (23%) lesions were obliterated with EVT only, DS after EVT, and DS

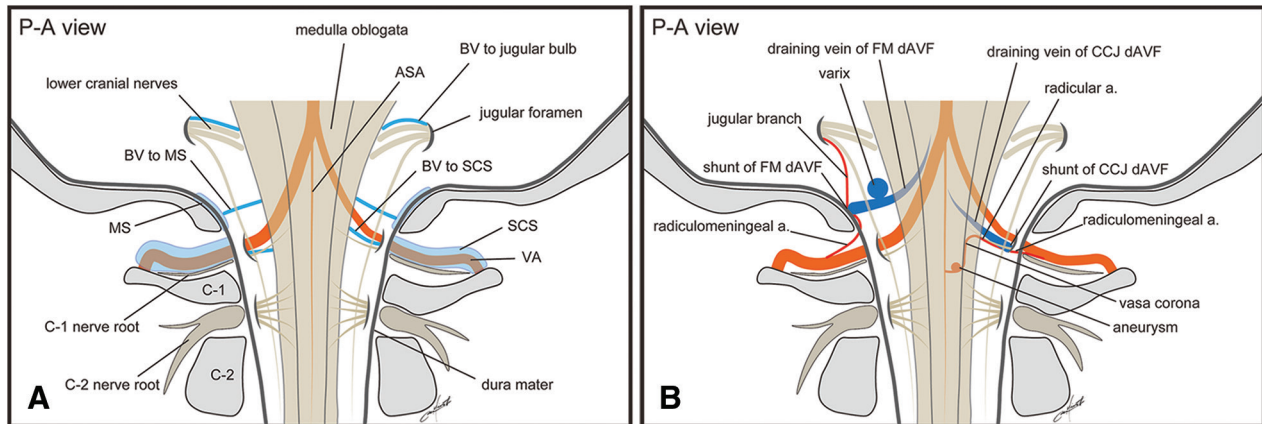


Fig. 2 (A) Schematic illustration of normal anatomy of the FM and CCJ. (B) Schematic illustration of vessel anatomy with special reference to the difference between the FM dAVF and CCJ dAVF. The FM dAVF is fed by the jugular branch and radiculomeningeal artery from the VA. The jugular branch is from the OA or APA. The shunt point of the FM dAVF is located on the dura mater at the FM. The FM dAVF may develop varix on the draining vein. The CCJ dAVF is fed by the radiculomeningeal artery and spinal pial arteries, such as the ASA and LSA. Vasa corona is the feeder from the spinal pial arteries and may develop an aneurysm on the feeder. The shunt point of the CCJ dAVF is located on the occiput-C1 level dura mater. APA: ascending pharyngeal artery; ASA: anterior spinal artery; BV: bridging vein; CCJ: craniocervical junction; dAVF: dural arteriovenous fistula; FM: foramen magnum; LSA: lateral spinal artery; MS: marginal sinus; OA: occipital artery; SCS: suboccipital cavernous sinus; VA: vertebral artery

only, respectively. Complete obliteration was obtained in 88% and 100% of lesions treated with EVT and DS, respectively. Complications occurred in three patients (7%) treated with EVT, which were ischemic complications, such as medulla oblongata infarction due to LSA occlusion,²⁷⁾ cerebellar infarction due to Onyx migration to the basilar artery,²⁶⁾ and lower cranial nerve palsy after embolization of stylomastoid artery.¹³⁾

Takai et al. reported a multicenter cohort study of the CCJ AVF, demonstrating that the DS was more effective and safer than the EVT.⁴⁴⁾ The retreatment rates after DS and EVT were 2.6% and 63%, respectively, and ischemic complication rates after DS and EVT were 7.7% and 26%, respectively. Yoo et al. reported the EVT results of medullary BV-draining dAVF comparing the FM and CCJ lesions.¹³⁾ In their series, the EVT resulted in complete or near-complete occlusion in all patients with FM dAVF and 61.5% of patients with CCJ AVF. The complication rates were 11.1% and 46.2% in FM dAVF and CCJ AVF, respectively. The higher occlusion rate and lower complication rate with EVT in FM dAVF, unlike CCJ AVF, may be due to less involvement of the spinal pial artery, such as ASA. Moreover, the external carotid artery (ECA) branches can be used as the target vessel of TAE, which is safer than the VA branches. However, we should recognize the possibility of anastomosis between the dural branch and spinal pial arteries during EVT.

Homology to the spinal dAVF

Mitsuhashi et al. reported the FM and CCJ dAVF as the dAVF draining into the BV of the medulla.¹¹⁾ They

speculated that these lesions are possible homologs of spinal dAVF and have similar characteristics. Regarding the CCJ AVF, spinal pial arteries may be involved, and the risk factors of hemorrhagic presentation are spinal artery involvement and feeder aneurysm.³⁸⁾ These characteristics are very different from spinal dAVFs at the thoracolumbar level.⁴⁵⁾ On the other hand, most FM dAVFs are supplied by dural arteries, and the cause of aggressive symptoms is the venous drainage pattern. Moreover, significant male predominance is similar to the spinal dAVF. The difference between the FM and spinal dAVF is hemorrhagic presentation. As Mitsuhashi et al. described, embryologically, the BVs of the medulla are cranial homologs of the spinal cord emissary BVs that drain the pial venous network.¹¹⁾ It seems reasonable to consider the FM dAVF as a cranial homolog of the spinal dAVF.

Conclusions

From the literature review, almost all of the feeders of FM dAVF were dural branches from VA, APA, and OA. Spinal pial arteries were rarely involved as the feeder. All lesions had venous reflux to the medulla oblongata via medullary BVs. The FM dAVF is characterized by a significant male predominance and a high incidence of aggressive symptoms. The most common symptom was congestive myelopathy, followed by hemorrhage. The FM dAVF differs from the CCJ AVF and is similar to the spinal dAVF. The EVT for the FM dAVF may be more effective and has lower

complication rates than that for the CCJ AVF. DS for the FM dAVF may be effective and safe.

Disclosure Statement

All authors have no conflict of interest regarding the present manuscript.

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