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TECHNICAL NOTE

Intraoperative DSA-guided minimal approach for craniocervical junction DAVFs obliteration

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Abstract

Background Dural arteriovenous fistulas (DAVFs) are a group of diseases involving problematic shunts between dural arteries and venous structures such as sinuses, meningeal veins, or even cortical veins. To focus on craniocervical junction dural arteriovenous fistulas (DAVFs), we introduce a minimally invasive technique with midline incision combined with intraoperative digital subtraction angiography (DSA). This hybrid technique can minimize the incision wound to an average of 6 cm which leads to less destruction and lower risk of adverse events.

Method Using this minimally invasive approach, surgical obliteration was achieved in 6 patients with craniocervical junction DAVFs. A minimal midline incision was made over the C1–2 level, measuring approximately 5 to 7 cm in length. C1 hemilaminectomy was performed for DAVF obliteration followed by intraoperative DSA for confirmation of complete obliteration.

Results Among these 6 patients, the radiculomedullary artery was the most common feeding artery. The mean length of the operation (including DSA performance) was 6.5 ± 1.4 h. None of these cases showed cerebrospinal fluid leakage or exacerbation of neurological symptoms after the operation.

Conclusion Using intraoperative DSA, the minimally invasive technique offers more precise but less destructive access than conventional far lateral suboccipital craniotomy. Most importantly, intraoperative DSA provided verification of complete closure for shunts that could not be examined for indocyanine green (ICG) dye because the microscope did not have a clear line of sight. In our experience, this technique shows encouraging results of fistula obliteration.

Keywords Dural arteriovenous fistula (DAVF) · Digital subtraction angiography (DSA) · Craniotomy · Indocyanine green (ICG) videoangiography · Hybrid operating room

Abbreviations

CTA	Computed tomography angiography
DAVFs	Dural arteriovenous fistulas
DSA	Digital subtraction angiography
ICG	Indocyanine green
MRA	Magnetic resonance angiography
SAH	Subarachnoid hemorrhage

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Introduction

Dural arteriovenous fistulas (DAVFs) are a group of diseases involving problematic shunts between dural arteries and venous structures such as sinuses, meningeal veins, or even cortical veins. These pathological shunts, most of which are idiopathic, account for 10–15% of intracranial arteriovenous malformations [16]. DAVFs at the craniocervical junction are uncommon but clinically important abnormalities. In contrast to thoracolumbar DAVFs, these lesions have a wide presentation, including acute subarachnoid hemorrhage [2, 6–8, 10, 11, 14, 18, 23], myelopathy [3, 4, 9, 13, 21, 24, 27, 33, 34, 36, 38], brain stem dysfunction [15, 29, 37], and even cranial nerve palsy [17]. Furthermore, DAVFs at the craniocervical junction are most common in middle age, and male patients outnumber female patients 3:1 [39].

To focus on craniocervical junction DAVFs, we introduce a minimal invasive technique with midline incision combined with intraoperative digital subtraction angiography (DSA). Compared with conventional far lateral approach for craniocervical junction DAVFs obliteration, this hybrid technique can minimize the incision wound to an average of 6 cm which leads to less destruction and lower risk of adverse events. On the other hand, visual obstruction caused by the retractor, other surgical equipment, and even nerve rootlets might mask indocyanine green (ICG) dye during videoangiography. Manipulation, such as retraction or excision of the rootlets for visualization, might result in neurological deficits. With the assistance of intraoperative DSA, verification of complete closure for shunts can be achieved.

Method

In this article, 6 patients with craniocervical junction DAVFs were recruited. Demographic data of these 6 patients were demonstrated in Table 1. All patients received operation with minimal midline incision combined with intraoperative digital subtraction angiography (DSA). Postoperative magnetic resonance angiography (MRA) was performed 64.2 ± 61.4 days after the operation to confirm that the pathological shunt was completely occluded.

Surgical technique

The neurosurgical dissections were performed in the hybrid operating room using a radiolucent carbon skull clamp set (DORO RADIOLUCENT skull clamp set, PRO MED Instrumente GmbH, Germany) and a TRUMPF operating table (TRUMPF Medizin Systeme GmbH + Co., Saalfeld, Germany) to simulate realistic surgical conditions. A standard operating microscope (OPMI, Carl Zeiss Company, Oberkochen, Germany) was employed.

First, we prepared an endovascular access route via the femoral artery with a long guiding sheath for later intraoperative DSA in the supine position. For the subsequent craniocervical junction operation in the prone position, two-thirds of the length of the guiding catheter was kept out of the femoral artery and positioned lateral to the upper thigh. Then, the patient was placed in a prone position with the head fixed in a radiolucent carbon head frame. Sterilization and draping procedures were carried out. The hub of the sheath was also sterilized and draped for intraoperative use (Fig. 1).

Second, a minimal midline incision was made over the C1–2 level, measuring approximately 5 to 7 cm in length; the position was determined by marking the spinous process of C2. Monopolar electrocautery was used to incise the subcutaneous fat and then deepen the incision in the avascular plane of the ligamentum nuchae. An expandable tubular retractor system (Spine Classics, Aesculap AG, Germany) was applied to maintain the surgical field and trajectory. The retractor was further expanded laterally to obtain a better surgical corridor by ipsilateral paraspinal

Table 1 Demographic data, clinical presentation, fistula characteristics, mRS scores, and operation data

Patient	1	2	3	4	5	6
Age	70	54	71	75	65	62
Sex	M	F	M	M	M	M
Clinical presentation	Sudden-onset headache	Persistent headache for 4 days	Sudden-onset change in consciousness	Intermittent headache for 6 months	Progressive paraplegia	Severe dizziness
GCS at admission	E3V5M6	E4V5M6	E1VeM1	E4V5M6	E4V5M6	E4V5M6
Feeding arteries	C1 RA of VA; OA	C1 RA of VA	C1 RA of VA	C1–3 RA of VA	C1 RA of VA	C1 RA of VA
Drainage direction	Rostral	Both rostral and caudal	Rostral	Rostral	Both rostral and caudal	Both rostral and caudal
mRS score	3	0	5	4	5	0
Angiographic result	Complete obliteration	Complete obliteration	Complete obliteration	Complete obliteration	Complete obliteration	Complete obliteration
Operation time	8 h	6 h	8 h	7.5 h	5 h	5 h
Intraoperative blood loss	450 mL	500 mL	600 mL	630 mL	350 mL	150 mL

GCS Glasgow Coma Scale, *RA* radicular artery, *VA* vertebral artery, *OA* occipital artery, *mRS* modified Rankin Scale

Fig. 1 Intraoperative DSA preparation and patient positioning. The upper panel shows preparation for endovascular access via the left femoral artery with a long guiding sheath for later intraoperative DSA; the lower panel demonstrates the prone position for subsequent DAVF obliteration surgery

Fig. 2 The minimal incision along the midline was suitable for the placement of an expandable retractor (Spine Classics, Aesculap AG, Germany). The upper panel displays a 5-cm midline incision made for the approach; the lower panel shows our expandable retractor that we utilized in the operation

muscle dissection on the ipsilesional side of the lateral mass of C1, and then hemi-laminectomy of C1 was performed (Figs. 2 and 3).

Third, the dissection was extended cranially to the ipsilateral craniocervical junction alongside the foramen magnum. Part of the median condyle was ground away with a drill to enlarge the foramen magnum ipsilaterally. A paramedian dural mater incision was performed, following sharp dissection of the denticulate ligaments. After lateral and ventral dissection of the arachnoid membrane, an abnormal engorged venous structure was revealed (Fig. 3). After identification of one or several fistula points (usually found near the C1 rootlet where the vertebral artery enters the intradural space), complete obliteration was performed by clipping; success was verified intraoperatively with ICG dye.

Finally, intraoperative DSA was performed. By consulting the three-dimensional reconstruction image, we identified the spatial relationship between the clips and the tortuous vascular structures. Most importantly, intraoperative DSA provided verification of complete closure for shunts that could not be examined for ICG dye because the microscope did not have a clear line of sight.

Results

We managed 6 patients with craniocervical junction DAVFs during the study period. Ethics approval was not required for this study. Informed consent has been obtained according to institutional guidelines. All patients except one presented with subarachnoid hemorrhage (SAH), and the remaining patient presented with congestive myelopathy. The 6 patients included 5 men and 1 woman with a mean ± standard deviation age of 66.7± 7.5 years. Among these 6 patients, the radiculomedullary artery was the most common feeding artery; the occipital artery acted as an additional feeding artery in 1 patient. Three patients had drainage in the rostral direction only, and 3 patients had both rostral and caudal drainage. Most patients showed relatively good neurological status on hospital admission; however, 1 patient showed poor neurological status, with a Glasgow Coma Scale E1VeM1. The clinical characteristics of this series are summarized in Table 1.

All 6 patients received immediate surgery, and the mean length of the operation (including DSA performance) was 6.5 ± 1.4 h. The mean volume of intraoperative blood loss was 446.7 ± 177.3 mL. None of our surgical cases showed

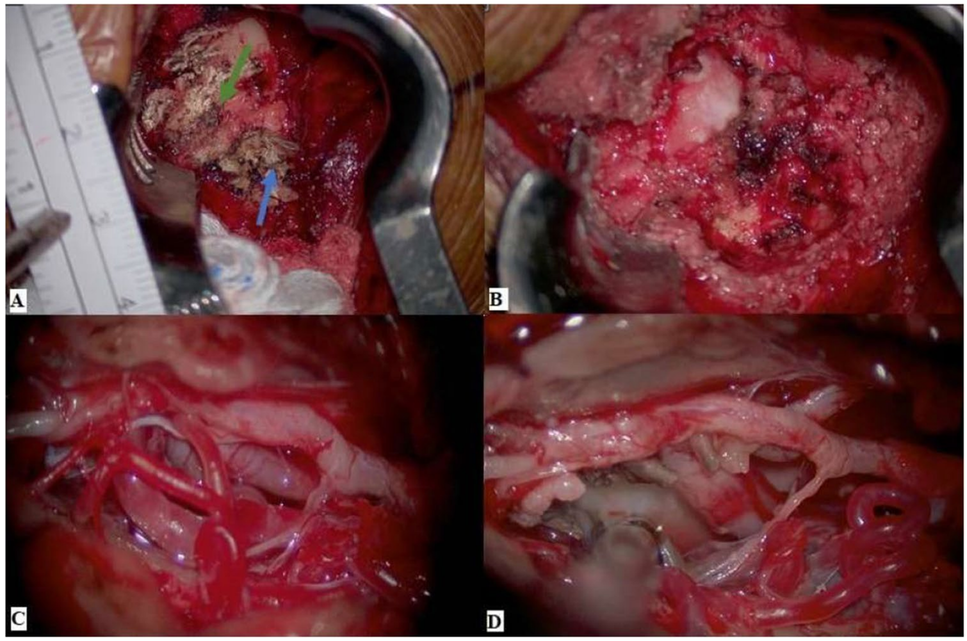

Fig. 3 Intraoperative photographs. (**A**) Use of a retractor for hemilaminectomy of C1 (green arrow indicates the occipital condyle; blue arrow indicates the posterior arch of C1). (**B**) Removal of the posterior arch of C1 and part of the occipital condyle. (**C**) Engorged and tortuous vessels were noted after the denticulate ligament was cut. (**D**) The pathological vessels were excised after coagulation

cerebrospinal fluid leakage or exacerbation of neurological symptoms after the operation, and their mean modified Rankin Scale score was 2.8 ± 2.3. Postoperative magnetic resonance angiography (MRA) was performed 64.2 ± 61.4 days after the operation to confirm that the pathological shunt was completely occluded.

Case illustration

Case 1

A 52-year-old female patient suffered from intermittent headache for 6 months. She came to our neurological clinic for help. The neurological examination showed no obvious abnormal findings. Computed tomography angiography (CTA) showed engorged vessels over the anterior aspect of the foramen magnum and anterior spinal vein of the upper cervical spine (Fig. 4A, B). Under the impression of craniocervical junction DAVF, surgery for lesion obliteration was performed via a midline minimal-incision approach. Intraoperative DSA revealed a DAVF located at the right margin of the foramen magnum with some minute feeding arteries from the meningeal branch of the right vertebral artery at the C1 level (Fig. 4C, D). The pathological shunting arteries were coagulated and excised. The patient achieved marked symptom relief without any adverse events postoperatively.

Case 2

A 63-year-old male patient suffered from a sudden onset of severe dizziness and was brought to the emergency department. Initial brain CT was performed, revealing subarachnoid hemorrhage in the prepontine cistern and hydrocephalus. Craniocervical junction DAVF was found on further DSA examination. Then, the patient received DAVF obliteration surgery with a midline minimal-incision suboccipital approach. With the assistance of intraoperative DSA, pathological shunts were soon recognized (Fig. 5A). During microsurgery for DAVF coagulation, ICG videoangiography was utilized to check whether these pathological vessels were eliminated. A feeding artery was initially missed; fortunately, however, it was detected by the second intraoperative DSA (Fig. 5B). This small, ventrally located artery was coagulated (Fig. 5C, D). The patient's postoperative recovery was smooth, without any new-onset neurological deficits. Subsequent brain MRA confirmed that the vascular lesions were no longer present.

Discussion

At present, the optimal treatment strategy for craniocervical junction DAVFs remains controversial. Both surgical obliteration and endovascular techniques (including transvenous or transarterial embolization) face difficulty because of the deep-seated lesion locations and complicated angioarchitecture. Compared with thoracolumbar lesions, fistulae in the craniocervical region are relatively hazardous to access surgically. Nevertheless, Takami et al. [34] proposed a microsurgical technique that directly interrupts retrograde arterial flow of blood in the medullary vein to the spinal cord; this modality is safe and effective. Sorenson

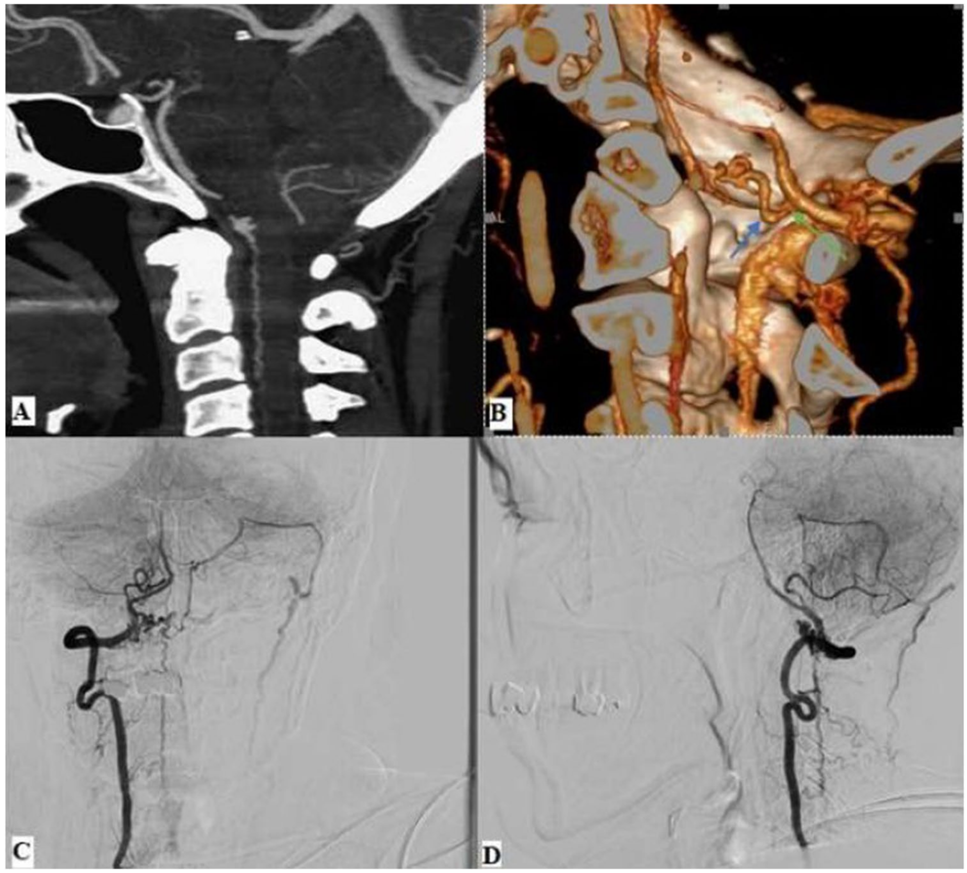

Fig. 4 Radiological findings showing craniocervical junction DAVF with tortuous venous dilatation in a 54-year-old female patient. (**A**) Computed tomography angiography (CTA) shows engorged vessels over the anterior aspect of the foramen magnum and anterior spinal vein. (**B**) The blue arrow indicates the engorged drainage vein, while the green arrow indicates the fistula shunting point. (**C** and **D**) Intraoperative DSA reveals a dural AV fistula at the right margin of the foramen magnum with some minute feeding arteries from the meningeal branch of the right vertebral artery at the C1 level

et al. [31] also showed a case of successful clip ligation of a symptomatic craniocervical junction DAVF that had previously undergone attempted Onyx embolization. Although endovascular treatment of DAVFs seems to be less invasive, its feasibility is largely dependent on the angioarchitecture of the lesion [35]. The risk of infarction of arteries feeding critical brainstem structures, especially if the fistula has multiple tortuous feeders, makes endovascular therapy an inappropriate option [25]. It is speculated that venous hypertension from embolic agents at shunting fistulae or potential injury and thrombosis in the sinus might play a critical role in DAVF recurrence [12]. Regarding treatment results, Steinmetz et al. [32] performed a meta-analysis that showed an obliteration rate of 98% with microsurgery compared to 46% with embolization. In our opinion, surgical intervention is the most acceptable and effective modality to treat DAVFs at the craniocervical junction.

According to the literature, conventional surgical treatments for craniocervical junction DAVFs are based on far lateral suboccipital craniotomy or craniectomy [1, 30] For this approach, we used a “hockey stick” incision, which starts at the level of the superior nuchal line where the mastoid tip is located, curves to the midline, and then descends straight to the spinous process of the axis vertebra [28, 35]. Muscle splitting and dissection from the insertion at the superior nuchal line to the spinous process of the second vertebra are necessary to obtain a better surgical field, which leads to postoperative neck pain and dysesthesia [5]. In addition to the above complications, the far lateral approach is more likely than the midline minimal-incision approach to face the dilemma of cerebrospinal fluid leakage because a wider piece of the dura is excised [22]. To overcome the shortcoming of postoperative nuchal pain, Rawanduzy et al. proposed a modified far-lateral approach with more minimal linear incision [26], avoiding the excess tissue disruption resulted from an L-shaped incision that split muscle over the transverse process of C1.

In our experience, a midline ipsilaterally minimal-incision technique can minimize soft tissue destruction via the hybrid utilization of microsurgery and intraoperative DSA. As reflected by our experiences in a hybrid operating room [19], intraoperative DSA can provide an encouraging level of accuracy in minimally invasive surgical DAVF ligation. Therefore, we can minimize the incision wound to an average of 6 cm, which leads to less destruction and blood loss than conventional far lateral suboccipital craniotomy. Owing to the minimization of anatomical destruction, this approach can also reduce complications including

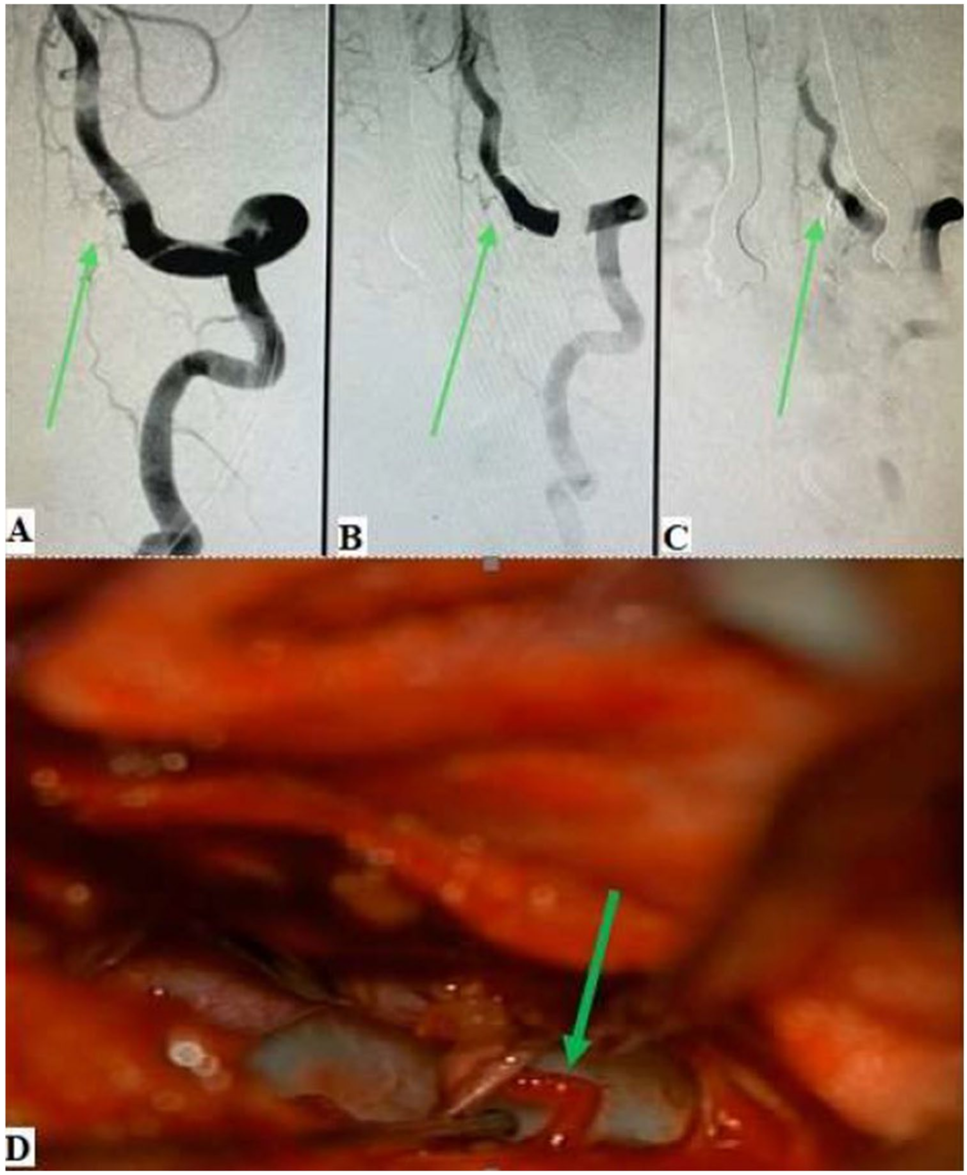

Fig. 5 Intraoperative DSA for confirmation of complete lesion obliteration. (**A**) First intraoperative DSA before obliteration of the DAVFs. (**B**) The second DSA revealed a minute residual feeding artery. (**C**) During the final DSA confirmation, a temporal clip was placed to rule out the possibility of a residual shunting fistula. (**D**) The minimal residual feeding artery was finally found under a microscope

postoperative nuchal pain, dysesthesia, and impaired wound healing. Compared with the far- lateral minimal linear incision from Rawanduzy et al., a relatively familiar midline approach followed by ipsilaterally dural incision could make us clearly identify the intradural vertebral artery entry site and prevent catastrophic vascular damage. Furthermore, cerebrospinal fluid leakage can be prevented owing to the small size of the dural incision and the scarcity of space in which cerebrospinal fluid can accumulate. Most of all, our surgical experiences showed promising results in that all 6 patients achieved total obliteration of their fistulas.

Finally, we demonstrate a case (Fig. 5) with a small ventral feeding artery that was blocked from view by the retractor and must could not be seen during microscope assisted ICG videoangiography. Under the microscope, it was difficult to inspect the fine angioarchitecture because of the visual hindrance caused by the retractor, other surgical equipment, and even nerve rootlets. The visual obstruction also masked ICG dye during videoangiography. Manipulation, such as retraction or excision of the rootlets for visualization, might result in neurological deficits [20]. Fortunately, intraoperative DSA successfully located the residual lesion, and ligation was achieved. Complete obliteration of the shunting artery was subsequently confirmed by intraoperative DSA.

Limitations

However, there are still some limitations concerning this novel surgical technique. First, the surgeon needs to adapt to the smaller surgical field that familiarity of the anatomical structure of the craniocervical junction is necessary. In addition, skill of the microsurgical dissection, hemostasis, and duroplasty would be more difficult for those less experienced surgeons. Another limitation is the setup and time cost which makes some surgeons considering the intraoperative DSA is overdoing. However, we think that craniocervical junction DAVFs are relatively rare and complicated vascular malformations that certain precision and least operative risk are obligatory.

Conclusion

Surgical treatments for craniocervical junction DAVFs are always complicated and challenging due to the deep-seated locations and complicated angioarchitecture of these lesions. Through the use of intraoperative DSA, the minimally invasive technique offers more precise access than conventional far lateral suboccipital craniotomy. In our experience, this novel technique shows an encouraging rate of fistula obliteration and ensures that no residual shunts are missed.

Authors' contributions All authors contributed to the study conception and design. In this manuscript, CR and CHL wrote the main manuscript. YST and CCS revised the text and prepared the figure description. All authors read and approved the final manuscript.

Declarations

Ethics approval and consent to participate This article does not contain any studies with human participants performed by any of the authors.

Competing interests The authors declare no competing interests.

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Comments

The authors describe a minimally invasive approach to treat dAVF of the craniocervical junction. They use intraoperative DSA and then a midline incision to treat the fistulous connection where the vertebral artery pierces the dura at C1. They use a bivalve speculum for retraction. Overall, this is an interesting approach using the retractor system to use a muscle splitting technique. They treated 6 patients with this technique successfully. They verified successful treatment of the craniocervical dAVF by intraoperative DSA. The necessity of use of DSA is not clear if they use ICG following ligation.

The idea was interesting to use a muscle splitting technique from the midline. As an alternative, our group prefers to achieve a minimally invasive approach using a far lateral approach through a direct lateral incision centered over the transverse process of C1 [1].

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