# Management of craniocervical junction arteriovenous fistula fed by the radicular and spinal arteries draining into a common intradural vein : A case report

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# Introduction

Arteriovenous fistulas at the craniocervical junction (CCJ AVF) are rare vascular malformations, accounting for less than 2% of patients with intracranial or spinal AVFs; their most common presentation is subarachnoid hemorrhage.<sup>1-3</sup> Though a precise understanding of the angioarchitecture of CCJ AVF is essential for appropriate treatment and achieving curative treatment, preoperative definitive diagnosis can be difficult due to its complexity, and intraoperative findings may provide clues to the definitive diagnosis. Here, we report a case of a radicular CCJ AVF fed by the radicular and spinal arteries draining into a common intradural vein, which was finally diagnosed based on intraoperative findings and treated by disconnecting the common draining vein.

# **Case presentation**

A 57-year-old male presented with sudden cervical pain due to a subarachnoid hemorrhage, predominantly in the posterior cranial fossa. Neuroradiological examination revealed an AVF at the CCJ with the right C2 radicular and spinal pial arteries branching from the anterior spinal artery (ASA) as feeders, which drained into the anterior spinal vein via a single intradural vein. Fusion images of computed tomography angiography and magnetic resonance cisternography revealed that the fistula was located along the anterior root of the C2 nerve. Since the preoperative images could not identify the exact shunt point of the feeder from the spinal pial arteries branching from the ASA, a preoperative diagnosis of radicular AVF, concurrent dural and perimedullary AVF, and dural AVF with a pial feeder was made. Direct surgery was planned because the AVF was fed by the branches of the ASA and endovascular treatment was considered to increase the risk of spinal infarction. Small right suboccipital craniotomy, C1 laminectomy, and partial C2 laminectomy were performed. During surgery, an AVF was detected in the C2 anterior roots, as shown in the preoperative neuroradiological findings. Therefore, we made a diagnosis of radicular AVF rather than dural or perimedullary AVF. A common single drainer was disconnected surgically, and the disappearance of the shunt flow was confirmed using intraoperative digital subtraction angiography and indocyanine green videoangiography. The postoperative course was uneventful, and the patient returned to normal life.



Figure 1. Microsurgical anatomy of the craniocervical junction A and B) Posterior views of the microsurgical anatomy of the craniocervical junction.





Figure 4. Source images CT angiography of the and fusion images of the CT angiography and magnetic resonance cisternography. A-D) These images revealed that the shunt point seemed to be located in the intradural space and close to the right C2 anterior nerve root. Wide black arrow, shunt point.

Operation

# **Preoperative Neuroradiological Examinations**



#### Figure 2. Preoperative neuroradiological examinations.

A) Computed tomography (CT) images showed a subarachnoid hemorrhage predominantly in the posterior cranial fossa. B) Posterior view of the CT angiography image revealed abnormal vessels in the craniocervical junction (area marked by dotted lines).





D





Figure 3. Preoperative vertebral artery angiography (VAG). A) Preoperative early arterial phase of the anteroposterior view of the right VAG showed an arteriovenous fistula of the craniocervical junction fed by the C2 radicular artery. B) Preoperative late arterial phase of the anteroposterior view and p arterial phase of the lateral view of right VAG (C) showed the arteriovenous fistula has ascending intradural drainage through the anterior spinal vein and epidural drainage. D) Enlarged view of B. E) Preoperative early arterial phase of the anteroposterior view of the left VAG revealed that the same arteriovenous fistula was fed by the branches from anterior spinal artery. F) Preoperative late arterial phase of the anteroposterior view of the left VAG showed the same arteriovenous fistula of the craniocervical junction drained into the anterior spinal vein. G) A schematic drawing of the vascular structure. Asterisk, fistula; Black arrowhead, anterior spinal artery; White arrowheads, anterior spinal vein; White wide arrow, right C2 radicular artery; White narrow arrow, vertebral venous plexus; Ant., anterior; A., artery; Vert., vertebral; Rt., right; Lt., left



#### Figure 5. Intraoperative images.

Shunt point has been exposed from rostral side (A) of the C2 nerve, between the C2 nerve roots (B), and caudal to the C2 nerve (C). D) Intraoperative ICG videoangigraphy image. A single draining vein has been disconnected. The inlet showed the direction of the operative view. QR code, operative video

# **Postoperative angiography**



### Figure 6. Pre- and **Postoperative vertebral artery** angiography

Preoperative right (A) and left (B) anteroposterior view of the vertebral artery angiography. Postoperative right (C) and left (D) anteroposterior view of the vertebral artery angiography. These images showed complete obliteration of the fistula while preserving the anterior spinal artery.

#### References

- 1. Hiramatsu, M., Sugiu, K., Ishiguro, T., Kiyosue, H., Sato, K., Takai, K., Niimi, Y., & Matsumaru, Y. (2018). Angioarchitecture of arteriovenous fistulas at the craniocervical junction: a multicenter cohort study of 54 patients. Journal of Neurosurgery JNS, 128(6), 1839-1849.
- 2. Song, Z., Ma, Y., Wang, Y., He, C., Li, G., Zhang, P., ... & Zhang, H. (2023). Arteriovenous fistulas in the craniocervical junction region: with vs. without spinal arterial feeders. Frontiers in Surgery, 9, 1076549.
- 3. Ma, Y., Song, Z., Wang, Y., Wang, J., He, C., Li, G., ... & Zhang, H. (2024). Clinical features, treatment strategies and outcomes of craniocervical junction arteriovenous fistulas: a cohort study of 193 patients. Stroke and Vascular Neurology, 9(1).

## Discussion

This case indicates that a CCJ AVF with multiple feeders draining into a common intradural draining vein can be successfully treated by disconnecting the vein. CCJ AVFs often exhibit complex angioarchitecture. Understanding the angioarchitecture of the lesion using various modalities and planning appropriate treatment strategies are essential for their treatment.

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