



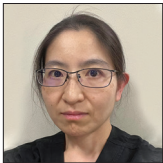
Case Report

# Intracranial venous reflux without the central venous occlusive disease in a patient receiving hemodialysis through brachio-brachial arteriovenous fistula: A case report

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## ABSTRACT

**Background:** Upper-limb arteriovenous fistula as a hemodialysis access among patients with end-stage renal disease (ESRD) has become a preferred type of vascular access. However, complications involving the central nervous system may occur. There have been no reported cases of internal jugular vein (IJV) regurgitation without central venous occlusive diseases (CVODs). We describe the case of a patient on HD who presented with symptomatic IJV regurgitation without CVODs.

**Case Description:** An 83-year-old man with ESRD receiving HD through a left upper-limb AVF presented with impaired consciousness and seizures. After recovery from unconsciousness, he became alert with cognitive impairment. The left subclavian arteriography revealed early filling of the left subclavian vein due to the AVF on the left brachium, with retrograde high-flow venous reflux to the left IJV, sigmoid and transverse sinuses, with the left central veins patent. All cerebral venous drainage procedures were dependent on the right IJV. The left internal carotid arteriography showed venous congestion of the left hemisphere. The flow of the left brachial artery was measured extremely high. Under compression of the left brachial artery to reduce the flow, the regurgitation persisted. With the findings that all cerebral venous return were in the right IJV, sacrificing the left IJV was thought to be acceptable. Left IJV ligation was performed, and the patient's cognitive function improved.

**Conclusion:** The short-term outcome after IJV ligation may be positive in the patient who was confirmed to have a normal cerebral venous return route independent of the refluxed IJV.

**Keywords:** Central venous disease, End-stage renal disease, Internal jugular vein valve, Internal jugular vein, Pseudophebitic pattern

## INTRODUCTION

The number of patients with end-stage renal disease (ESRD) undergoing hemodialysis (HD) has been increasing worldwide. HD access remains a crucial aspect of patient management, especially concerning quality of life without complications. Patients undergoing HD require vascular access through an arteriovenous fistula (AVF) or arteriovenous graft to obtain an adequate blood flow

rate to perform HD if they are independent of a central venous catheter.<sup>[14]</sup> Despite known changes in hemodynamics and cardiac remodeling associated with AVF, it remains the preferred type of vascular access.<sup>[15]</sup>

Complications involving the central nervous system may occur and there have been many reports of internal jugular vein (IJV) regurgitation in HD patients with the central venous occlusive diseases (CVODs).<sup>[1,2,4,8,9,11-13,16-20,22-27]</sup> To the best of our knowledge, there have been no reported cases of IJV regurgitation without CVODs.

Here, we describe the case of a patient on HD who presented with symptomatic IJV regurgitation without CVODs. We also discuss the etiology of IJV regurgitation and treatment of cerebral venous hypertension due to IJV regurgitation.

The patient provided informed consent to publish this case report and his identity was protected.

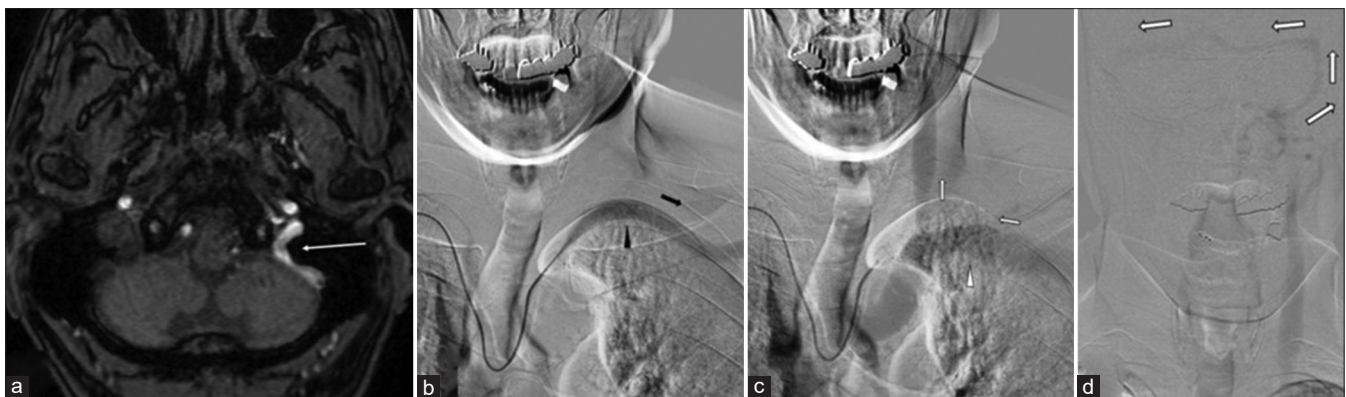
## CASE DESCRIPTION

In this report, we present the case of an 83-year-old right-handed male patient with ESRD. The patient, who had been receiving HD for 1.5 years through a left upper-limb AVF, was brought to our hospital on the day following his last HD due to impaired consciousness and status epilepticus. The patient's left arm was not edematous. His medical history included chronic renal failure, ischemic coronary artery disease, and post craniotomy state from the total removal of meningioma on the left frontal convexity 20 years before admission. He independently drove himself to receive biweekly HD treatment before admission. After controlling his seizures using anticonvulsants, he underwent imaging studies, including brain computed tomography and magnetic resonance (MR) imaging/angiography. MR angiography showed a high signal in the right transverse sinus, sigmoid sinus, and jugular bulb [Figure 1a], which indicated a

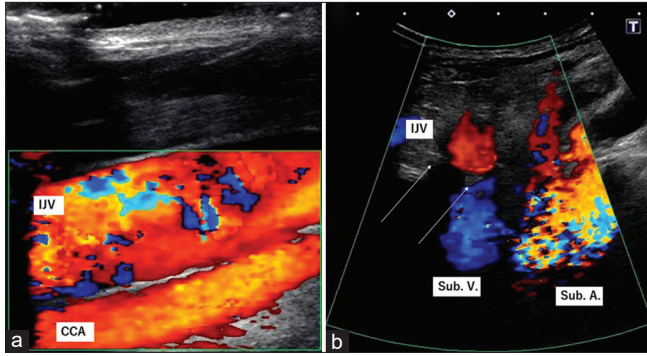
suspected dural AVF. MR imaging revealed a postoperative lesion at the left frontal convexity, which had been known for 20 years with no other significant findings.

The patient experienced aphasia and right hemiparesis for 2 days after gaining consciousness. The Mini-Mental State Examination (MMSE) was used to evaluate the patients' cognitive function immediately after he became alert, and a score of 16 points was measured, indicating moderate cognitive impairment. Digital subtraction angiography (DSA) was performed and left subclavian arteriography (AG) revealed early filling of the left subclavian vein due to the AVF on the left brachium, with retrograde high-flow venous reflux to the left IJV, sigmoid sinus, and transverse sinus, with the left brachiocephalic vein and superior vena cava patent. The retrograde venous flow was inserted into the confluence and drained through the right IJV [Figures 1b-d]. All cerebral venous drainage procedures were dependent on the right IJV. The left internal carotid AG showed delayed venous flow and drainage with venous congestion of the left superior middle cerebral vein as well as stagnant flow in the left transverse-sigmoid sinus junction. Based on these findings, we concluded that the patient's seizure was caused by venous hypertension in the left cerebral hemisphere due to IJV regurgitation induced by the upper-limb high-flow AVF.

Further examinations were performed using duplex ultrasonography (US). The flow of the left brachial artery was measured at 3150 ml/min, which indicated that the extremely high flow was produced by the brachio-brachial AVF. The brachial vein bifurcated into two major trunks on the proximal side of the anastomosis. Retrograde venous flow to the left IJV from the left subclavian vein was revealed using US [Figure 2a]. The left IJV valve was then visualized [Figure 2b]. The regurgitation increased during the Valsalva maneuver. The left brachial artery was compressed to reduce



**Figure 1:** MR angiography at admission showing a high signal in the left jugular bulb (arrow in a). The left subclavian arteriography showing the left subclavian artery (black arrowhead of b) flowing distally (black arrow in b), the left subclavian vein (white arrowhead in c) flowing into the left brachiocephalic vein with the retrograde flow into the left internal jugular vein, the left sigmoid sinus, and transverse sinus, inserting to the confluence, and draining through the right transverse sinus (white arrows in c and d).

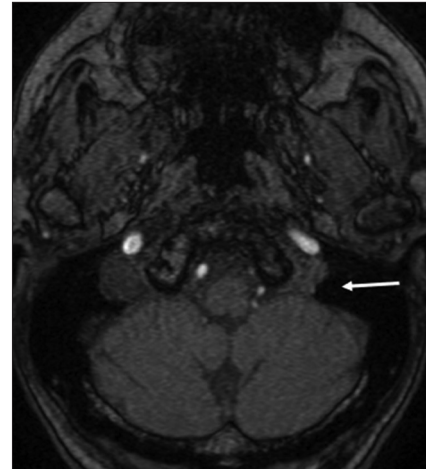


**Figure 2:** Duplex ultrasonography before surgery showing regurgitation in the left internal jugular vein (a), and visualizing internal jugular vein valves (arrows in b). IJV: internal jugular vein, CCA: common carotid artery, Sub. V.: subclavian vein, Sub. A.: subclavian artery.

the flow to 600 ml/min, but the regurgitation persisted. When the flow was decreased to zero, there was no regurgitation to the IJV, but this was not an acceptable solution for optimal blood circulation to the left upper limb. The patient also underwent cardiac US and coronary angiography and was diagnosed with Forrester Type I heart failure, which does not require surgical treatment.

Vascular surgeons along with cardiovascular physicians and neurosurgeons discussed the treatment strategy. A flow reduction procedure of the left brachio-brachial AVF was suggested for long-term cardiac outcome under HD, with prevention of heart failure by a high-flow shunt; however, this procedure was not sufficient to terminate the left IJV regurgitation caused by the high-flow AVF. There was no need for an endovascular approach to the central veins because they were patent. With the finding that all cerebral venous return was in the right IJV, the sacrificing the left IJV was thought to be acceptable. The first priority was to terminate IJV regurgitation to urgently normalize intracranial pressure, followed by flow-reduction surgery of the AVF for a long-term systemic prognosis.

Thus, the left IJV ligation was performed under general anesthesia. Videoangiography using indocyanine green performed before ligation revealed retrograde venous flow in the left IJV. Videoangiography performed after ligation showed no flow into the IJV. Immediately, after surgery, the patient's MMSE score improved to 24 points and persisted until discharge. Post-operative US showed no regurgitation to the ligated IJV, with high flow of the brachial artery. Brain MR angiography did not show high-intensity signals in any of the intracranial dural sinuses [Figure 3] with no new lesions in the brain. After confirming successful termination of intracranial venous reflux, the patient underwent surgery for AVF flow reduction. A short interposition with a small-caliber prosthetic graft was performed on one arterialized



**Figure 3:** MR angiography after surgery showing the disappearance of a high signal in the left jugular bulb (arrow).

main trunk of the brachial vein and the other trunk was ligated, which successfully reduced the left brachial artery flow to 920 ml/min, as well as spared persistent AVF as a good vascular access route. The postsurgical course was good and the patient was independently discharged.

## DISCUSSION

The well-known adverse effects of the upper-limb AVF for HD include left ventricular hypertrophy, high-output cardiac failure, exacerbation of coronary ischemia, and central vein stenosis.<sup>[15]</sup> High-flow AVF might cause complications including steal syndrome, venous hypertension, and aneurysmal degeneration in addition to those mentioned above.<sup>[10]</sup>

In most cases, the IJV valve, which is reportedly the only venous valve between the right atrium of the heart and the brain, has been thought to play an important role in preventing regurgitation from the subclavian vein to the IJV, especially when intrathoracic pressure is increased. However, acquired or congenital IJV valve incompetence may impair cerebral venous return under the Valsalva maneuver resulting from IJV regurgitation affecting cerebral circulation.<sup>[3,5-7,11]</sup> Some HD patients with the upper-limb AVF might have chronic venous hypertension of the brachial and/or subclavian veins. Anatomical studies have shown that chronic venous hypertension causes IJV valve dysfunction.<sup>[11]</sup> However, the procedure for judging IJV valve competency remains controversial.<sup>[3,21]</sup>

Cases of intracranial venous reflux associated with CVODs in HD patients with neurological deficits have been reported.<sup>[1,2,4,8,9,11-13,16-20,22-27]</sup> One of the assessed mechanisms was the coexistence of venous hypertension with CVODs and incompetent IJV valves. However, after thoroughly



inspecting the literature, we found no reported cases of IJV regurgitation without CVODs.

Reported treatments for regurgitation due to high-flow upper-limb AVF include termination of the AVF (ligation or occlusion), angioplasty with or without stenting of stenotic central veins, and anticoagulant therapy; the last two preserve the AVF.<sup>[1]</sup>

Our patient reportedly had IJV regurgitation due to an extensive high brachio-brachial AVF flow, without CVOD. The IJV valve was visible to the patient. IJV regurgitation observed in the supine position increased under the Valsalva maneuver, suggesting IJV valve incompetence. The flow of the left brachial artery was excessive (up to 3150 ml/min). In contrast to previously reported cases, the IJV regurgitation in our patient was thought to be caused by IJV valve incompetence and excessive AVF flow, not by CVOD. The findings suggested that compressing the brachial artery to achieve optimal blood flow did not prevent regurgitation. DSA revealed that all cerebral venous outlets utilized the right IJV. Ligation of the left IJV was the treatment implemented to achieve preservation of functional AVF and termination of IJV regurgitation. This treatment may have been the best course of action for this patient as it urgently reduced intracranial pressure, resulting in good neurological outcomes.

This study had limitations as it was a case report that may have incidentally shown a positive clinical outcome. The patient had not been observed for a long period in terms of general condition, including cardiac outcome and AVF patency, as well as neurological condition.

## CONCLUSION

We report a case of symptomatic cerebral venous reflux due to IJV regurgitation without CVOD. IJV regurgitation without CVOD is rare. The short-term outcome after IJV ligation may be positive in the patient who was confirmed to have a normal cerebral venous return route independent of the refluxed IJV after thorough observation. Long-term follow-up and further accumulation of cases are crucial to confirm the relevance of these treatment results.

## DECLARATIONS

### Ethics approval and consent to participate

Not applicable.

### Availability of data and materials

The data that support the findings of this case are available on reasonable request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

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