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Case Report

Endovascular Treatment of Cerebral Venous Circulation Dysfunction Caused by Hemodialysis Shunt: A Case Report

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Objective: Central venous disease, defined as ≥50% stenosis or obstruction of central veins, is one of many life-threatening complications faced by patients on hemodialysis. It often presents as upper limb edema to the arteriovenous (AV) shunt for hemodialysis, although neurological symptoms are rare. We report a case of central venous disease with neurological symptoms associated with endovascular therapy.

Case Presentation: A 79-year-old man presented with status epilepticus. His past medical history included rectal carcinoma when he was 69 years old and indication for hemodialysis when he was 79 years old. However, he had no history of neurological disease or epilepsy. On arrival at our facility, CT perfusion revealed venous circulation dysfunction on the left cerebral hemisphere. DSA demonstrated regurgitation from the AV shunt on left upper limb to the cerebral veins and obstruction of the left subclavian vein. Ligation of the causal AV shunt was deemed difficult due to surrounding edema; therefore, endovascular transarterial coil embolization was performed. After completely occluding the AV shunt, patient's condition improved significantly. The patient was discharged 3 days later without neurologic symptoms, with no recurrence of epilepsy was observed to date.

Conclusion: Coil embolization of causal AV shunt significantly improved the neurological symptoms of central venous disease.

Keywords central venous disease, hemodialysis, cerebral venous circulation

Introduction

Central venous disease (CVD), defined as ≥50% stenosis or obstruction of central veins (either the superior and inferior vena cava, brachiocephalic vein [BCV], subclavian vein [SCV], or internal jugular vein [IJV]),[1)] has been considered one of the many life-threatening complications observed among hemodialysis patients, with its incidence having been estimated to range from 16% to 50%.[1)] CVD often presents as upper limb edema ipsilateral to the hemodialysis shunt, with neurologic symptoms rarely occurring.[1,2)]

We herein report a case with cerebral venous circulation dysfunction caused by hemodialysis shunt and compare our findings with those published previously.

Case Presentation

Our case involved a 79-year-old man who had no history of neurological disease. At 69 years, the patient had undergone surgery and chemotherapy due to rectal carcinoma, during which a central venous catheter (CVC) had been placed in his left SCV. After 4 years, the CVC had been removed due to catheter-related blood stream infection and subsequently transferred into his right SVC, which was later removed within the same year. By the time he was 78 years old, an arteriovenous (AV) shunt had been placed on his left upper limb for indication of hemodialysis. However, another AV shunt had been placed on the right upper limb soon after due to left upper limb edema. Around 1 month before admission, he was diagnosed with lung carcinoma. Moreover, his family noticed that he was having seizures, for which he was transported to our facility.

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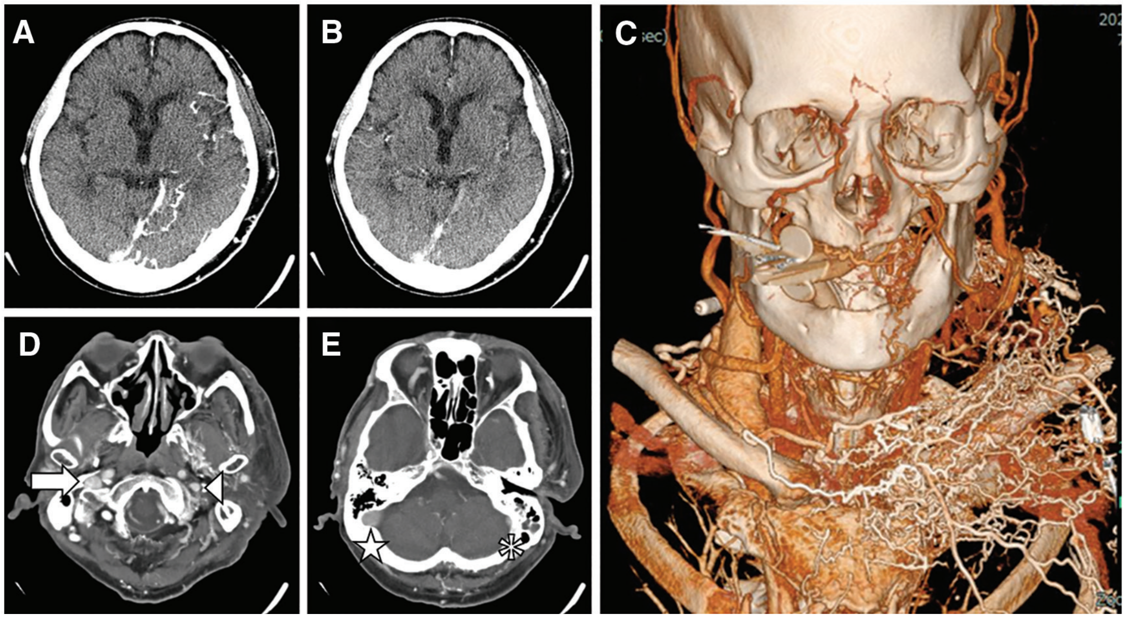

Fig. 1 Contrast CT images. In early phase of CTP source image, cerebral veins of left hemisphere were enhanced (**A**). In late phase of CTP source image, cerebral arteries were enhanced (**B**). In 3D CTA, both right IJV and superficial veins of chest wall were enhanced but left IJV was not enhanced (**C**). In source images of 3D CTA, compared with right IJV (arrow), left IJV (arrowhead) was hypoplastic (**D**). Similarly, left sigmoid sinus (asterisk) was hypoplastic compared with right sigmoid sinus (star) (**E**). CTP: CT perfusion; IJV: internal jugular vein

On arrival, the patient still had a seizure with a Japan Coma Scale (JCS) of III-200. Based on the diagnosis of status epilepticus, the patient was provided anti-seizure medication intravenously and subsequently intubated. Although non-contrast CT showed no abnormality, source image of CT perfusion (CTP) showed enhancement of the cerebral veins followed by the cerebral arteries (**Fig. 1A** and **1B**). In 3D CTA, superficial veins of left chest wall and right IJV were enhanced, but not the left IJV (**Fig. 1C**). Furthermore, source images of 3D CTA demonstrated hypoplasia of left IJV and left sigmoid sinus (**Fig. 1D** and **1E**). To establish a diagnosis, DSA was performed.

First, the contrast agent was injected from the left peripheral vein of the upper limb, which revealed venous blood flow regurgitating into the cerebral veins via the left EJV and flowing into the right IJV (**Fig. 2A** and **2B**). Left subclavian arteriography revealed that blood flow from the AV shunt on the left upper limb regurgitated into the deep cerebral veins and superior sagittal sinus (SSS), and that the left SCV was obstructed (**Fig. 2C**–**2E**). The left transverse sinus (TSS) and left sigmoid sinus were not enhanced on right carotid arteriography (**Fig. 2F**) and left carotid arteriography showed similar finding (**Fig. 2G**). Based on these findings, we surmised that regurgitation from the left AV shunt into veins was diverted into cerebral veins due to left SCV obstruction and that such abnormal blood flow caused status epilepticus.

Should such dysfunction of cerebral venous circulation persist, the patient would have been at risk for severe complications, such as intracranial hemorrhage. Although obstruction of the left upper limb AV shunt was considered, ligation was deemed implausible given the surrounding edema. Therefore, transcatheter arterial embolization (TAE) was indicated. Under general anesthesia, the right brachial artery was punctured, a 6-Fr short sheath was placed, and a long guiding catheter (ASAHI FUBUKI 6Fr AN/110cm; ASAHI INTECC, Aichi, Japan) was guided into the left subclavian artery. Excelsior SL-10 microcatheter (Stryker, Kalamanzoo, MI, USA) was placed into left radial artery and guided adjacent to the AV shunt. After inserting and detaching the coil, it immigrated to basilic vein through the AV shunt due to blood flow (**Fig. 3A**). Therefore, coil embolization was applied under left upper limb compression by hand (**Fig. 3B**). In total, 38 coils including AXIUM 3D 14 mm × 40 cm (Medtronic, Dublin, Ireland), AXIUM PRIME Frame 12 mm × 50 cm, Penumbra SMART COIL SOFT 8 mm × 20 cm (Medico's Hirata, Tokyo, Japan), Penumbra SMART COIL EXTRA SOFT 4 mm × 10 cm, and Penumbra SMART COIL WAVE EXTRA SOFT 3 mm × 6 cm were deployed to complete the shunt occlusion (**Fig. 3C**). The puncture site was closed using ANGIO SEAL VIP 6 Fr (Terumo, Tokyo, Japan).

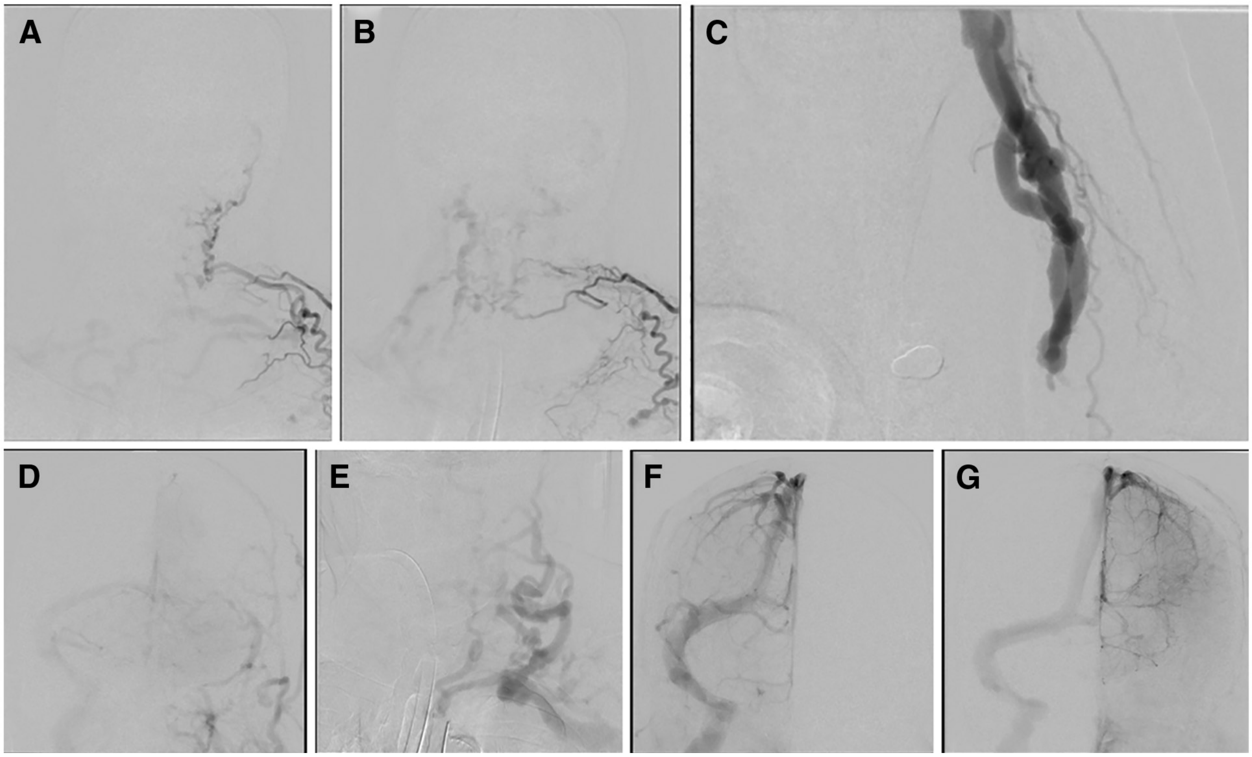

Fig. 2 Findings of DSA. Peripheral venography of left upper limb revealed regurgitation into cerebral veins via left EJV (**A**), and outflow to right IJV was observed (**B**). Left subclavian arteriography revealed that blood flow from the left upper limb AV shunt regurgitated into the cerebral veins (**C** and **D**) and that the left SCV was obstructed (**E**). In right carotid arteriography, left TSS and left sigmoid sinus were not enhanced (**F**). Left carotid arteriography showed similar finding (**G**). AV: arteriovenous; EJV: external jugular vein; IJV: internal jugular vein; SCV: subclavian vein

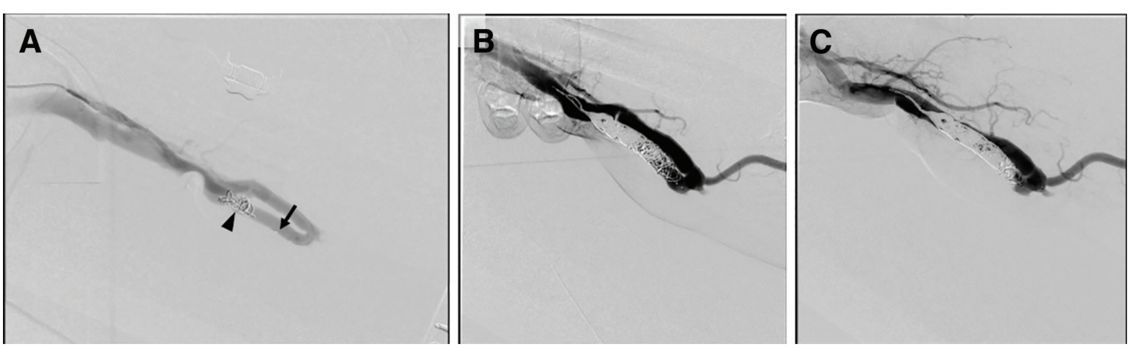

Fig. 3 TAE against the AV shunt of left upper limb. First coil was deployed from the microcatheter (arrow), but after detachment, the coil (arrowhead) immigrated into basilic vein due to blood flow (**A**). Therefore, coil embolization was performed under compression by hand (**B**). After deploying 38 coils in total, the AV shunt was completely occluded (**C**). AV: arteriovenous; TAE: transcatheter arterial embolization

After TAE, the patient was admitted to our facility. Around an hour after TAE, the patient's JCS improved to I–3, and spontaneous breathing and oxygenation were stable. Therefore, the patient was weaned from mechanical ventilation and extubated. Given the patient suffered no recurrence of epilepsy or neurological symptoms, he was discharged on day 3. After discharge, the patient has been followed up at the outpatient department and continues to receive hemodialysis with an AV shunt on his right upper limb. Around 1 month after the discharge, CTP showed normal cerebral blood flow (**Fig. 4A**–**4D**). In 3D CTA, right IJV, right sigmoid sinus, right TSS, and right SSS were enhanced, but both left IJV and left TSS were not enhanced, with left sigmoid sinus barely enhanced (**Fig. 5A** and **5B**).

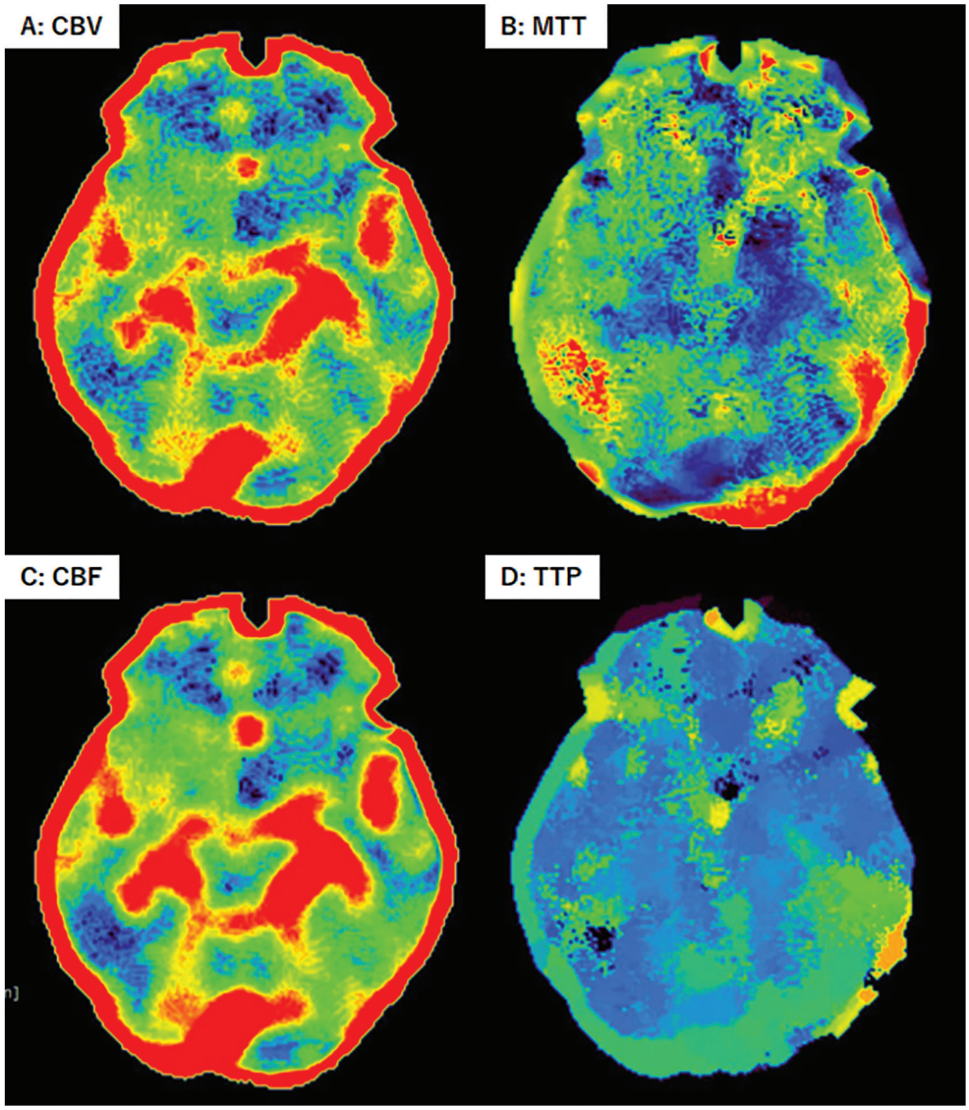

Fig. 4 CTP finding 1 month after the discharge. CBV (**A**), MTT (**B**), CBF (**C**), and TTP (**D**) showed normalized cerebral blood flow. CBF: cerebral blood flow; CBV: cerebral blood volume; CTP: CT perfusion; MTT: mean transit time; TTP: time to peak

Discussion

A literature review of case reports had shown that among 23 cases of CVD with neurological symptoms, 20 cases had obstruction of BCV and 6 cases had a history of CVC placement. Among those who had history of CVC placement, three cases underwent CVC placement into the SCV similar to the current case.[1)] CVC placement has been associated with an increased risk of CVD even after its removal, with studies suggesting that CVC placement into the SCV, prolonged placement, placement into the left side of the neck, and multiple catheter placement were especially associated with an increased risk for CVD.[1,3)] In our case, the patient underwent CVC placement into the left BCV for the treatment of rectal carcinoma around 10 years before the admission, which had been removed 4 years later. On admission, DSA revealed left BCV obstruction. Evidence has strongly suggested that prolonged CVC placement into the left BCV conferred to this obstruction, which promoted regurgitation of blood flow from the left upper limb AV shunt into the cerebral veins. In addition, findings of image studies on admission and 3D CTA 1 month after discharge suggested the hypoplasia of both left IJV and left sigmoid sinus, and aplasia of left TSS. In a case report of venous congestive encephalopathy due to right BCV occlusion and blood flow from AV graft on ipsilateral arm to IJV and cerebral veins, hypoplasia of left TSS was present.[4)] In both this case and the current case, abnormality of veins may have blocked venous outflow and accelerated cerebral venous congestion. In our case,

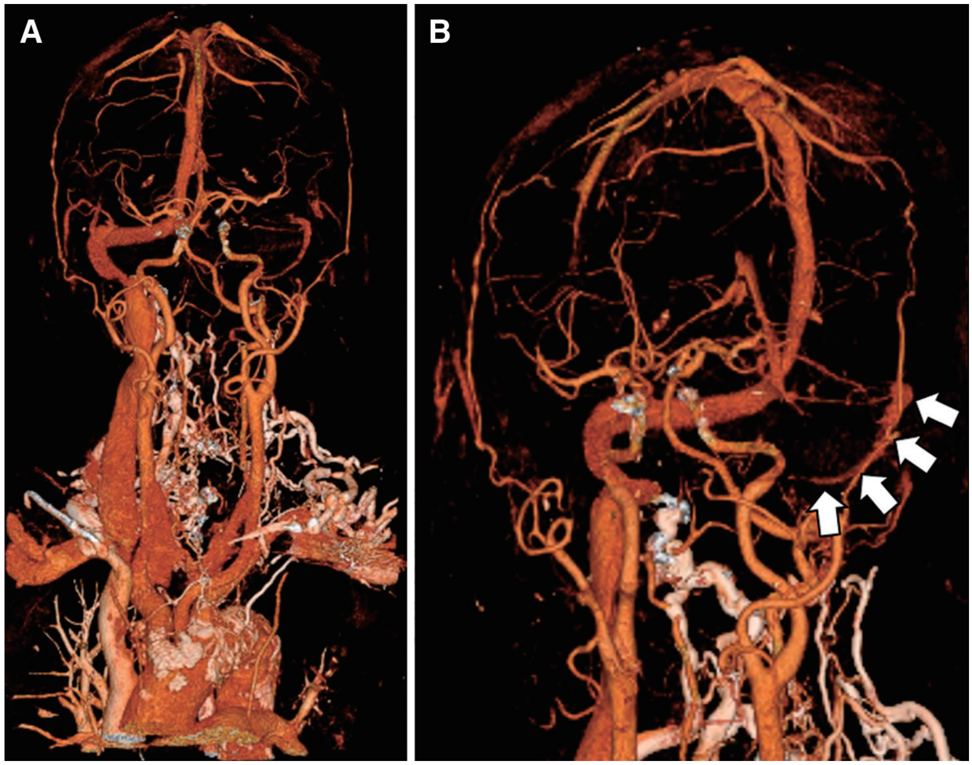

Fig. 5 3D CTA finding 1 month after the discharge. Right IJV, right sigmoid sinus, and right TSS and SSS were enhanced, but left IJV was not enhanced (**A**). Compared with right sigmoid sinus and right TSS, left sigmoid sinus (arrows) was barely enhanced and left TSS was not enhanced (**B**). IJV: internal jugular vein; SSS: superior sagittal sinus; TSS: transverse sinus

coil embolization of the AV shunt eliminated the regurgitation, after which rapid improvement of neurologic symptom was observed.

In the aforementioned literature review, 4 out of 23 cases with CVD had an occluded hemodialysis AV shunt.[1] In three cases, the AV shunt was occluded surgically,[2,4,5] whereas in one case, the AV shunt was occluded manually at the bedside.[6] Moreover, many of the other 19 cases were treated with AV shunt ligation or endovascular therapy for the BCV lesion (e.g., angioplasty and stent placement).[1,7–15] In our case, surgical ligation or obstruction was deemed implausible due to edema surrounding the AV shunt. Moreover, Eguchi and Honma reported that 69% of the case did not require reintervention for the original lesion 1 year after the stent placement against central venous lesion,[16] whereas Ashwal et al. also reported that 33% did not require recurrent stenosis or reintervention 1 year after the endovascular therapy (i.e., percutaneous transluminal angioplasty with or without stent placement) for central venous lesion.[17] Given these low patency rates, the approach reported herein may be one of the potential alternatives for intervention against symptomatic CVD. On the other hand, our approach has several limitations. First, embolization of hemodialysis AV shunt with coils for cerebrovascular purpose could be off-label use. Second, we inserted as many as 38 coils to occlude causal AV shunt because the microcatheter (i.e., Excelsior SL-10) was used. We could have used less coils if we had inserted a catheter with larger inner diameter and coils with larger secondary diameter, but there were no such coils in our facility. Moreover, immediate intervention was warranted in our case because of risk for severe complications, and therefore waiting for arrival of those devices was not a practical choice. Further discussion on cost-effectiveness is warranted for our approach. Third, if clinicians occlude the causal AV shunt, they subsequently need to make a new vascular access for hemodialysis. In our case, the patient already had another AV shunt on contralateral upper limb and therefore the patient could undergo annual hemodialysis soon after the intervention.

Similar to that reported in previous studies,[1,2] our case demonstrated upper limb edema ipsilateral to the causal AV shunt. In some cases, facial edema ipsilateral to the AV shunt has also been reported.[2,4] When such symptoms are observed after the placement of AV shunt for hemodialysis, clinicians may need to be concerned of the risk for cerebral venous circulation dysfunction. Moreover, when they are involved in the management of potential hemodialysis candidates, clinicians also need to be aware that CVC placement into BCV and prolonged CVC placement are associated with an increased risk for CVD.

Conclusion

We herein report a case of CVD with neurologic symptom who exhibited significant recovery after coil embolization of the causal hemodialysis AV shunt. Risk factors for CVD, such as prolonged CVC placement and CVC placement into BCV, need to be identified when clinicians are involved in the management of potential hemodialysis candidates.

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Disclosure Statement

All authors declare that they have no conflicts of interest.

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