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Neurological Symptoms Due to Intracranial Venous Congestion in a Hemodialysis Patient With Arteriovenous Shunted Flow

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INTRODUCTION

The incidence of strokes in hemodialysis patients is high, often leading to neurological disorders and a poor prognosis.[1] A stroke generally includes cerebral infarction and hemorrhage (intracerebral and/or subarachnoid hemorrhage).[1] The risk of cerebral hemorrhage during hemodialysis is high; previous studies have reported that the incidence of cerebral hemorrhages in hemodialysis patients was 8.7 per 1000 persons per year.[2] It is speculated that the main cause of cerebral hemorrhage in hemodialysis patients is hypertension, as the incidence of this condition during hemodialysis is relatively high.[1,2] Nonetheless, various other causes for the neurological disorder, such as cerebral aneurysm, cerebral arteriovenous malformation, and amyloid angiopathy, have been observed in prior reports.[3,4]

Hemodialysis patients require vascular access via an arteriovenous fistula (AVF) or arteriovenous graft (AVG) to obtain an adequate blood flow rate to perform hemodialysis. However, stenosis or obstruction of the outflow and/or central vein often causes venous congestion, resulting in edema of the affected upper limb.[5–7] In addition, albeit rare, the occurrence of venous congestive encephalopathy and subarachnoid hemorrhage has also been reported.[7,8]

Here, we report a case of cerebral hemorrhage due to intracranial venous congestion in a hemodialysis patient with an AVG in her left forearm. The intracranial venous congestion had developed via blood flow from the AVG, severe stenosis of the left brachiocephalic vein, and backward flow of the left internal jugular vein. We thus aim to discuss central venous stenosis, as well as to report the clinical course, features, and neurological prognosis of intracranial venous congestion in hemodialysis patients.

CASE PRESENTATION

The patient, a 73-year-old woman, presented with both facial and upper arm edema on her left side. She had previously initiated hemodialysis because of end-stage renal disease caused by chronic glomerulonephritis; in addition, although a living kidney transplantation was eventually performed, the deteriorating function of the transplanted kidney resulted in the re-initiation of hemodialysis. She had since undergone 14 years of maintenance hemodialysis. The procedures were performed via an AVG in her left forearm. She had sick sinus syndrome and had a cardiac pacemaker implanted via the right subclavian vein prior to her current episode. Medical history indicated asymptomatic cerebral infarction, for which she was prescribed aspirin, and her ascending aorta was replaced by an artificial vessel. In addition, she was frequently treated via percutaneous transluminal angioplasty for the anastomotic stenosis of the AVG and its outflow vein. Severe stenosis of the left brachiocephalic vein, as well as the backward flow of the left internal jugular vein, had been detected during a percutaneous transluminal angioplasty procedure (Figure 1). However, the reconstruction of the AVF or AVG in the opposite arm would be difficult because of the stenosis of the right subclavian vein; as the edema of her left face and arm had not worsened, the AVG in her left forearm remained in use. She was not treated with endovascular therapy because a re-stenosis of the left brachiocephalic vein

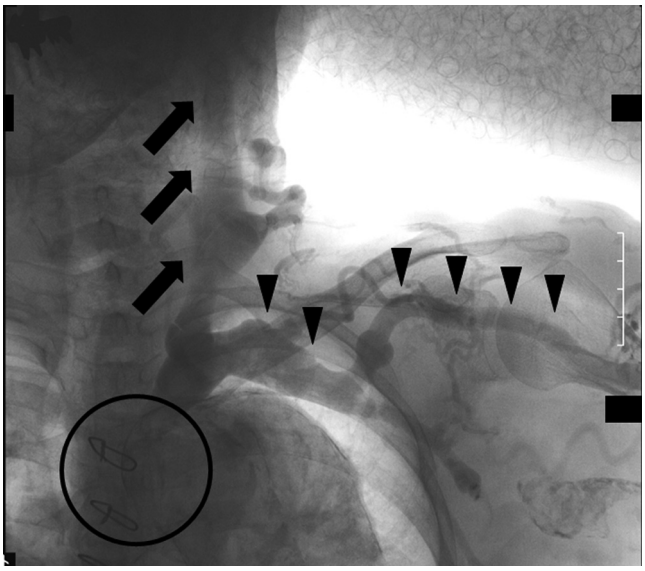

Figure 1. Angiogram of the arteriovenous graft in the patient's left forearm. Shunted blood flow from the arteriovenous graft in her left forearm (arrowheads). Severe stenosis of the left brachiocephalic vein (circle) and backward flow from the left internal jugular vein into the intracranial vein (arrows) were observed.

would probably have occurred after such a treatment. This speculation is supported by the findings of Lumsden *et al.*, which showed that the mean primary patency was 5.7 months in patients who were treated with endovascular treatment against central venous stenosis.[9] In addition, the patient did not want another repetition of endovascular therapy. Although the prominence of superficial veins was not observed, the small collateral superficial veins were greater on the left thoracic wall than on the right side. Two months before hospital admission, the patient noticed difficulty in speaking fluently, as well as the sudden onset of aphasia. In addition, a conversation could not be established; she presented with difficulty in speaking, verbal perseveration, paraphasia, disorientation, worsening of the left facial and upper limb edema, and left eye conjunctival hyperemia. Computed tomography revealed a high-density area in the left temporal lobe, indicating cerebral hemorrhage (Figure 2a); a widespread, high-density area was also detected in the left occipital lobe, indicating congestive edema (Figure 2b). We diagnosed venous rather than arterial bleeding as a result of the backward flow of the left internal jugular vein and a progression of the collateral circulation of the left intracranial vein; this was detected via a 4-dimensional computed tomogram (Supplementary Figure S1). We speculated that severe stenosis of the left brachiocephalic vein, combined with the reflux of the shunted blood flow into the left internal jugular vein, may have caused intracranial venous congestion, resulting in venous cerebral hemorrhage. We therefore performed an immediate operation to close the AVG. Two days after the procedure, disorientation, aphasia, and facial edema had improved, and left temporal lobe bleeding had decreased (Figure 3a and b). As for her vascular access, an operation established and subcutaneously repaired a superficial brachial artery in the right arm. After 1 month, she was discharged without any neurological sequelae.

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

DISCUSSION

In the current case, it was thought that the cerebral hemorrhage due to intracranial venous congestion was caused by central venous stenosis and blood flow from the AVG. Central venous catheter placement and cardiac pacemaker implantation are both known representative causes of central venous stenosis. Previous studies have reported that approximately 16% to 50% of central venous catheter placements can cause central venous stenosis.[5,7,9] Other causes, including radiation to the neck and chest area, tumor compression or invasion of the central veins, and deep vein thrombosis, have been also reported.[7] Patients who have developed severe kidney dysfunction often require the insertion of hemodialysis catheters into the central veins for emergency hemodialysis. Another study suggested that 11% to 40% of hemodialysis patients experience central venous stenosis[S1]; it is additionally thought that shunted blood flow may result in the proliferation of vascular endothelial cells in central veins, thus causing central venous stenosis.[7] Although previous reports suggest that almost half of hemodialysis patients who have AVFs or AVGs present with edema of the shunted limb,[5,7] it is rare for patients to develop neurological symptoms due to intracranial congestion and cerebral hemorrhage.[S2–S4] As observed in our patient, the collateral veins on the thoracic wall may be suggestive of central venous stenosis.[S5,S6] In addition, color Doppler ultrasonography may be useful to detect the backward flow of the intrajugular vein. Although color Doppler ultrasonography was not performed in the current case, Pereira *et al.* performed color Doppler ultrasonography of the intrajugular vein in a similar case and showed that the backward flow of the intrajugular vein remarkably decreased after the ligation of the arteriovenous fistula.[S2] Therefore, determining the existence and amount of the backward flow of the intrajugular vein evaluated by color Doppler ultrasonography can be a useful method for detecting an increase in the intracranial venous blood flow, resulting in intracranial venous hypertension.

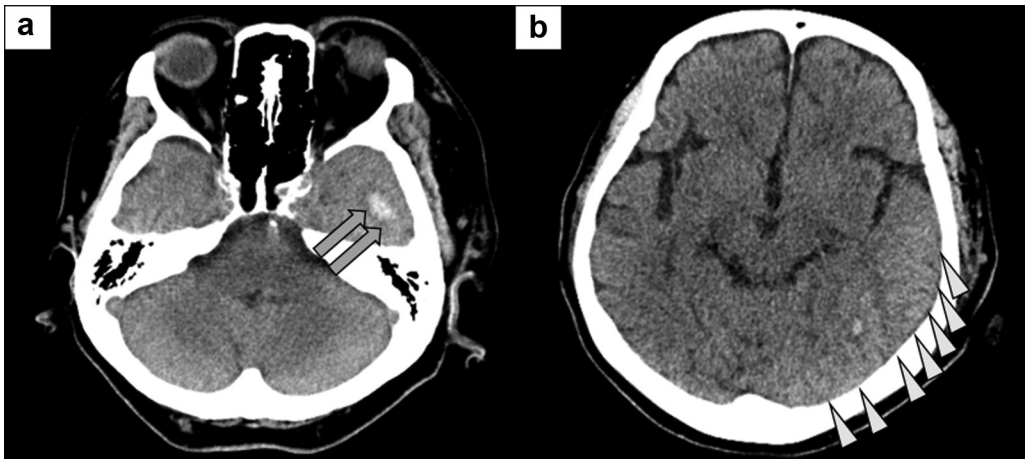

Figure 2. Plain computed tomogram of the head at the patients' hospital admission. There was (a) a high-density area in the left temporal lobe, indicating a cerebral hemorrhage (arrows). (b) In addition, (b) a widely spread, high-density area was detected in the left occipital lobe, indicating congestive edema (arrowheads); congestion of contrast agent in the left hemisphere demonstrated this finding.

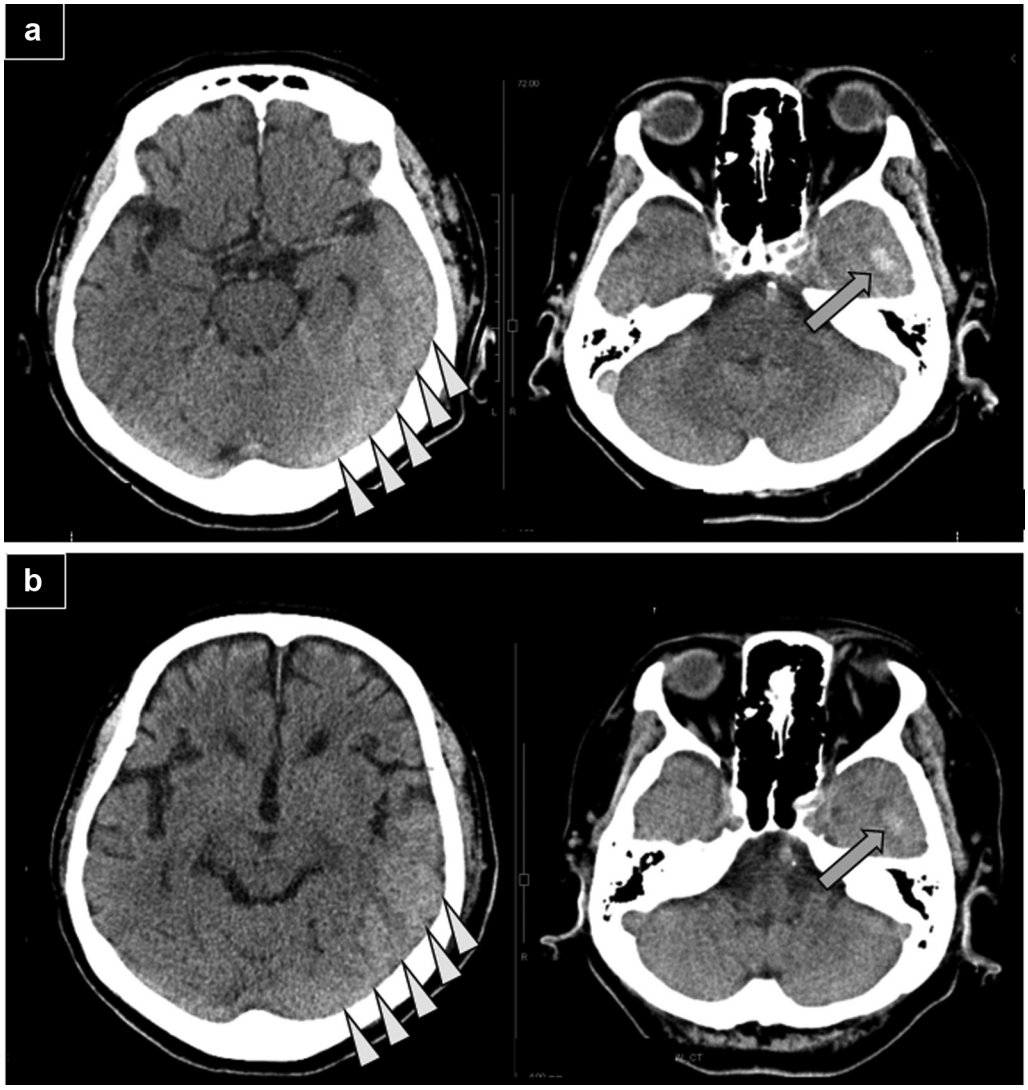

Figure 3. Plain computed tomogram of the patient's head after closure of the arteriovenous graft. One day (a) and 5 days (b) after arteriovenous graft closure. Left temporal lobe bleeding had gradually improved (arrows), as had the congestive edema caused by congestion of the contrast agent (arrowheads).

Table 1. Previous case reports of intracranial venous congestion in hemodialysis patients with arteriovenous shunt flow

References	Age, yr	Sex	Neurological symptoms	Neurological prognosis[a]
7	73	Male	Headache, disorientation, perseveration, blurred vision, seizure	Improvement in 2 d
7	67	Female	Disorientation, right arm weakness, jerking movement	Improvement
8	50	Male	Headache, agitation, disorientation, drowsiness, left homonymous hemianopia	Improvement in 1 wk
S2	43	Female	Headache, vertigo, left lateropulsion	Immediate improvement
S3	35	Female	Headache, ptosis, chemosis and exophthalmos in the right eye	Immediate improvement
S4	71	Female	Headache, nausea, vomiting	Improvement
S4	63	Female	Seizure	No intervention
S7	62	Male	Headache, transient loss of vision,	Improvement
S8	70	Male	Hononymous hemianopia, ataxia	No intervention
S9	74	Male	Loss of vision, blurred vision	Improvement
S10	59	Female	Headache, gait and stance instability, memory loss	Improvement after 1 wk
S11	28	Male	Ataxia, incoordination, vertigo, pre-syncope, fall towards left side	Improvement
S12	42	Male	Headache, sleepiness	Improvement in 1 d
S12	32	Male	Cognitive dullness, sleepiness	Improvement in 1 d
S13	13	Female	Headache, visual field defect, monocular blindness, seizure	Adequate intervention not performed
S14	42	Female	Loss of consciousness	Improvement
S15	47	Female	Headache, seizure, hemiplegia	Immediate improvement
S16	75	Female	Dizziness, seizure	Gradual improvement
S17	48	Female	Headache, nausea, blurred vision, photophobia	Immediate improvement
S18	69	Female	Left upper limb numbness and sensory abnormality, ataxia, incoordination	Improvement
S19	47	Male	Altered mental status, muscle weakness of right upper and lower extremity	Improvement in 1 wk
Current case	73	Female	Aphasia, paraphasia	Improvement in 2 d

[a]After the intervention.

Although there have been no other reports of intracranial venous congestion causing cerebral hemorrhage in hemodialysis patients, there are 22 cases (including our own) of intracranial venous congestion developing in hemodialysis patients with shunted peripheral arteriovenous flow (Table 1).[7,8,S2–S4,S7–S19] In these cases, the range of the patients' ages was between 13 and 75 years, and the male-to-female ratio was 9:13. Although the most common neurological symptom was headache, various symptoms were observed in these patients. The possibility of an arterial cerebral hemorrhage caused by hypertension, as well as intracranial venous congestion, should be considered in hemodialysis patients with shunted peripheral arteriovenous flow, particularly if the patient reports any neurological symptoms. Almost all patients exhibited improved symptoms within 1 week after an intervention to treat the congestion (performed via either endovascular treatment of the central vein or a closure of the arteriovenous shunt). In our case, although the patient developed a venous cerebral hemorrhage, she was discharged without any neurological sequelae. Thus, with the appropriate intervention, the neurological prognosis of symptomatic patients with intracranial venous congestion may be good and/or satisfactory. However, as Cleper *et al*. have reported, patients may develop repeated and prolonged convulsions following severe cerebral damage if the appropriate intervention is not performed.[S13]

In conclusion, the teaching points are summarized in Table 2. Most hemodialysis patients have AVFs or AVGs. The incidence of central venous stenosis is therefore high, as backward flow of the internal jugular vein and intracranial venous congestion may occur. In addition, there are various neurological symptoms caused by intracranial venous congestion. Thus, if hemodialysis patients develop a neurological disorder, the possibility of common cerebral vascular diseases, as well as intracranial venous congestion, should be considered. The neurological prognosis of such cases may prove promising despite complications associated with cerebral hemorrhage, as long as the appropriate therapeutic intervention is conducted.

Table 2. Teaching points

If a hemodialysis patient develops a neurological disorder, the possibility of common cerebral vascular diseases, as well as intracranial venous congestion, should be considered.
The neurological prognosis is good as long as an appropriate therapeutic intervention, such as an endovascular treatment of the central vein or a closure of the arteriovenous shunt, is performed.

DISCLOSURE

All the authors declared no competing interests.

SUPPLEMENTARY MATERIAL

Supplementary File (PDF)

Supplementary References.

Figure S1. Four-dimensional computed tomogram of the head. Contrast agent was injected into the peripheral vein of the right forearm. The right side of the intracranial veins

were enhanced, followed by the intracranial and collateral veins on the left; the left jugular vein was retrogradely enhanced.

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