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Case Report

Fatal Hemorrhage Immediately after Transvenous Embolization of Transverse-sigmoid Sinus Dural Arteriovenous Fistulas: A Case Report

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Objective: We report a case of intracerebral hemorrhage following emergency transvenous embolization for an acute symptomatic non-hemorrhagic dural arteriovenous fistula (dAVF).

Case Presentation: An 83-year-old woman demonstrated gait disorder and disturbance of consciousness. A transverse-sigmoid dAVF with retrograde deep venous drainage was detected on DSA. The left sigmoid sinus-jugular vein and the sinus confluence were occluded and the dAVF drains via the straight sinus (SS), medial superior cerebral veins and deep veins to the superior sagittal sinus (SSS). The dAVF was emergently treated by sinus packing of the transverse-sigmoid sinus with coils with contralateral approach via the occluded sinus confluence. Although the dAVF was markedly regressed, massive cerebral hemorrhage developed in the left parietal lobe immediately after embolization.

Conclusion: Although early treatment is required for dAVFs with aggressive symptoms, precious evaluation of their hemodynamics, particularly for drainage pattern, is mandatory to avoid a serious complication.

Keywords ▶ dural arteriovenous fistula, deep venous drainage, intracerebral hemorrhage, transvenous embolization

Introduction

In patients with Borden type 2 or 3 dural arteriovenous fistulae (dAVF) and non-hemorrhagic neurological symptoms, the annual incidence of hemorrhage is reportedly 10%, and in those with hemorrhage, it is reportedly 46%. In high-risk patients, curative interventions should be performed as early as possible to prevent hemorrhage. We report a patient with a non-hemorrhagic dAVF on detailed examination of rapidly progressing gait disorder and consciousness disturbance for whom diagnosis/emergency transvenous coil embolization were performed on the day of onset, leading to an effective decrease in shunt blood flow. However, extensive cerebral hemorrhage developed immediately after embolization probably due to insufficient assessment of drainage pattern of dAVF prior to treatment.

Case Presentation

Case: An 83-year-old female.

Complaints: Gait disorder and consciousness disturbance.

Present illness: She had been unemotional for the past 20 days. Gait disorder rapidly developed 4 hours before arrival, and she did not respond to words. She was bought to the emergency outpatient unit.

Medical history: Noncontributory.

Medication: Noncontributory.

Physical examination on arrival: Concerning the state of consciousness, the Japan Coma Scale (JCS) score was 30 and the Glasgow Coma Scale (GCS) score was E2V3M5. There was no response to calling, but there was no topical

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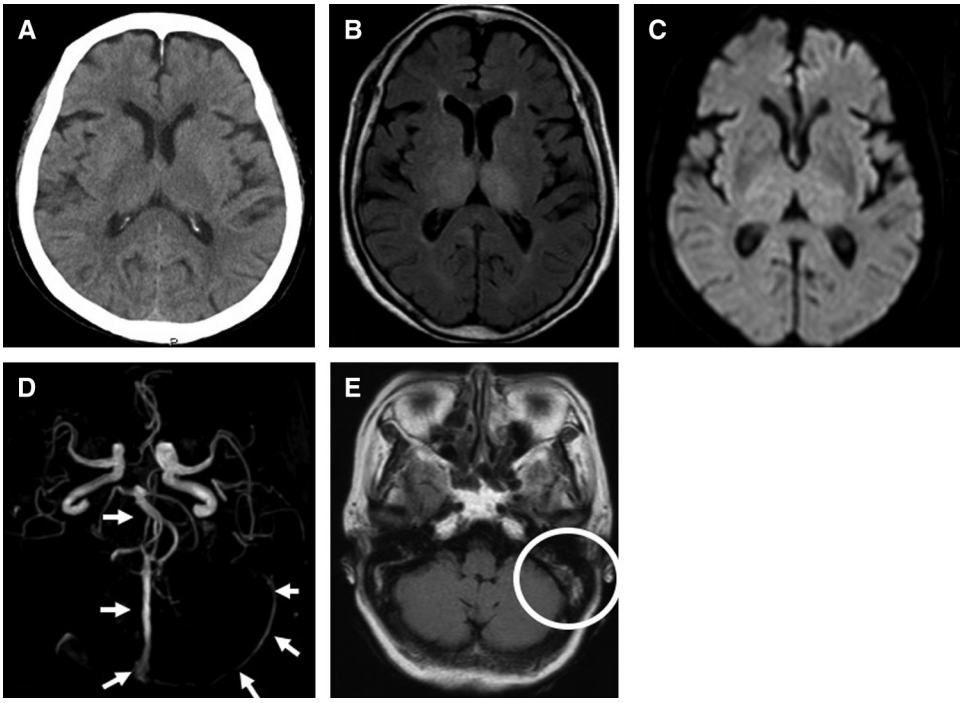

Fig. 1 (**A**) CT did not reveal cerebral hemorrhage. (**B**) FLAIR showed high-signal-intensity areas in the bilateral thalami. (**C**) Diffusion-weighted imaging did not show any changes in the signal intensity in the left or right thalami. (**D**) On MRA, the TS, SS, great cerebral vein of Galen, and internal cerebral vein were visualized with high signal intensity (arrow). (**E**) FLAIR showed high signal intensity in the left SigS, suggesting acute occlusion (round mark). SigS: sigmoid sinus; SS: straight sinus; TS: transverse sinus

neurological deficit, such as paralysis of the limbs, based on pain stimulation.

Hematology: Noncontributory.

Brain CT: Small low-density areas were detected in the bilateral thalami, but there was no cerebral infarction or hemorrhage.

Brain MRI: Acute cerebral infarction was not observed, but the bilateral thalami exhibited high signal intensity on FLAIR. In addition, the left sigmoid sinus (SigS) had high signal intensity, suggesting acute occlusion. Neither T2-weighted imaging nor susceptibility-weighted imaging (SWI) was conducted. On MRA, the straight sinus (SS), great cerebral vein of Galen, and internal cerebral vein were visualized with high signal intensity, suggesting dAVF draining retrogradely into the SS (**Fig. 1**).

DSA: DSA revealed a dAVF, with several shunt points at the left SigS and transverse sinus (TS), fed by the left occipital artery (OA), left ascending pharyngeal artery, and left middle meningeal artery. The left internal jugular vein was occluded and there was no antegrade blood flow from the shunt points. There was no communication with the sinus confluence, and SS-mediated reflux to the great cerebral vein of Galen, internal cerebral vein, septal vein, thalamostriate vein, cortical vein of parietal lobe, and the superior sagittal sinus (SSS) was observed. The flow of SSS, right TS, SigS, and internal jugular vein was antegrade, and the other draining route was left pterygoid venous plexus via the cavernous sinus (**Fig. 2**). Because A type III aortic arch made cannulation difficult, external carotid angiography was not performed. DSA demonstrated the congestion of the parietal lobe; deep vein congestion/cortical vein reflux may have increased the intracranial pressure, inducing neurological deficits such as gait disorder and consciousness disturbance.

Endovascular treatment: As an acute, symptomatic dAVF was complicated by gait disorder/consciousness disturbance, which were possibly associated with an increase in the intracranial pressure related to deep vein congestion/cortical vein reflux, emergency endovascular treatment was selected following DSA. Although transarterial embolization was also considered, it was difficult to access the fistula site due to type III aortic arch; therefore, transvenous embolization was performed in advance to promptly reduce congestion of the deep venous system. Because the left internal jugular vein was occluded, access was

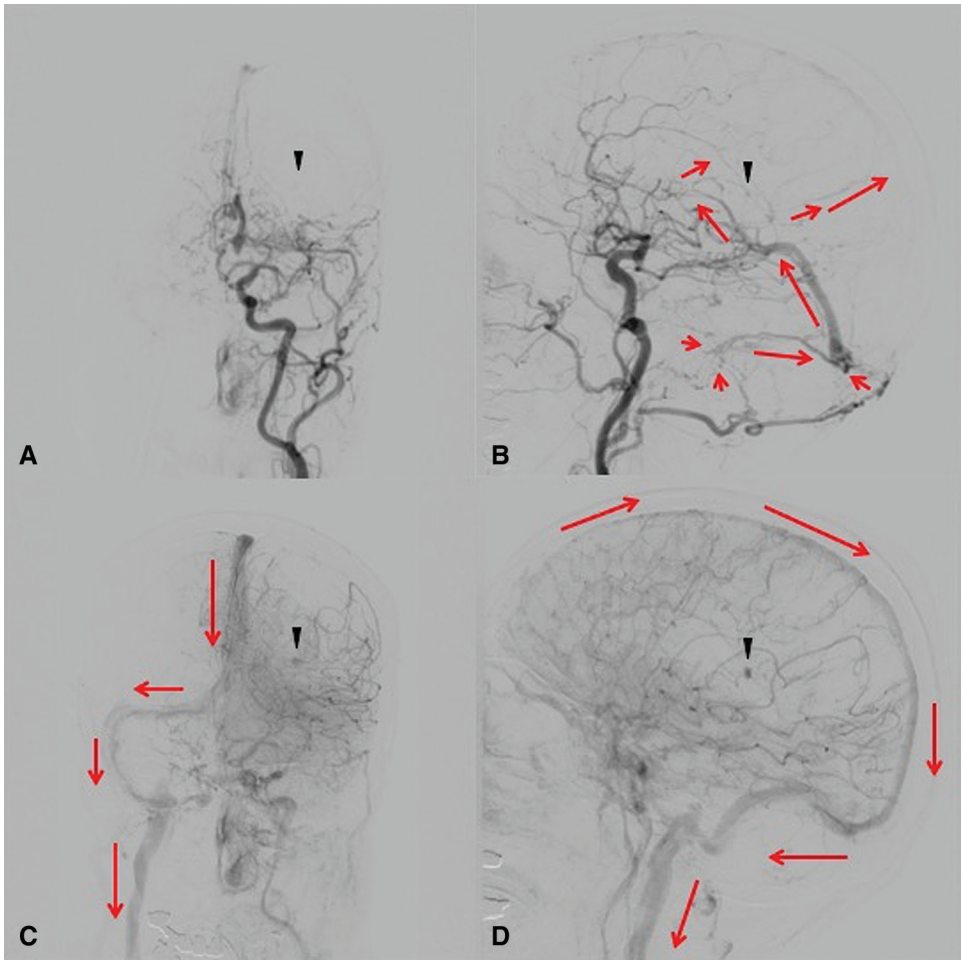

Fig. 2 (**A**) Frontal view on left common carotid angiography. Arterial phase. (**B**) Lateral view on left common carotid angiography. Arterial phase. (**C**) Frontal view on left common carotid angiography. Venous phase. (**D**) Lateral view on left common carotid angiography. Venous phase. After general anesthesia was induced, a guiding catheter was inserted and DSA was performed. DSA revealed a dAVF, with several shunt points at the left SigS and TS, fed by the left OA mainly. The left internal jugular vein was occluded and there was no antegrade blood flow from the shunt points. There was no communication with the sinus confluence, and SS-mediated reflux to the great cerebral vein of Galen, internal cerebral vein, septal vein, thalamostriate vein, cortical vein of parietal lobe, and the SSS (**B**: arrows) was observed. The flow of SSS, right TS, SigS, internal jugular vein was antegrade (**C**, **D**: arrows), and the other draining route was left pterygoid venous plexus via the cavernous sinus. DSA demonstrated the congestion of the parietal lobe. In the deep area of the parietal lobe, leakage of contrast medium was noted (arrowhead). dAVF: dural arteriovenous fistulae; OA: occipital artery; SigS: sigmoid sinus; SS: straight sinus; SSS: superior sagittal sinus; TS: transverse sinus

considered to be difficult. We planned sinus packing of the transverse-sigmoid sinus with coils with contralateral approach via the occluded sinus confluence. The patient was restless and it was difficult for her to remain still; therefore, this procedure was performed under general anesthesia. Considering the risk of hemorrhage, heparin was not administered during surgery.

As a diagnostic catheter, a 4Fr catheter (SY4; Gadelius Medical, Tokyo, Japan) was inserted into the left common carotid artery.

After inserting a sheath into the right femoral vein, an 8Fr catheter 80 cm (Brite tip; Cardinal Health Japan, Tokyo, Japan) was guided into the right internal jugular vein. At this point, extravascular leakage was observed in the deep area of the left cerebral hemisphere (**Fig. 2**).

As intermediate catheters, a 6Fr catheter (Cerulean DD6; Medikit, Tokyo, Japan) and 4.2Fr catheter (ASAHI FUBUKI; Asahi Intecc, Aichi, Japan) were guided to the left TS via the sinus confluence through the right TS. A microcatheter (Headway 17; Terumo, Tokyo, Japan) and

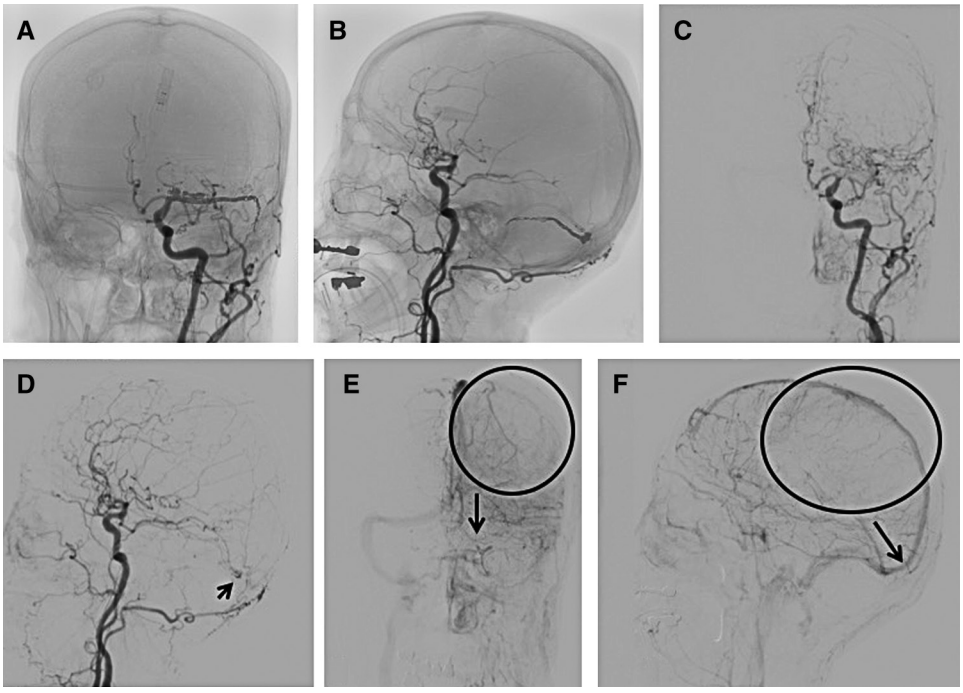

Fig. 3 (**A**) Frontal view on common carotid angiography after transvenous coil embolization. Arterial phase (DA). (**B**) Lateral view on left common carotid angiography. Arterial phase (DA). (**C**) Frontal view on common carotid angiography. Arterial phase (DSA). (**D**): Lateral view on left common carotid angiography. Arterial phase (DSA). (**E**) Frontal view on common carotid angiography. Venous phase (DSA). (**F**) Lateral view on left common carotid angiography. Venous phase (DSA). Coil embolization of the occluded end of the left SigS to left TS was performed. Although a small volume of shunt blood directly flowing into an area adjacent to the sinus confluence remained (**D**: arrow), antegrade outflow from the sinus confluence to the right TS was noted and reflux to the SS had disappeared; antegrade blood flow at the SS was observed (**E**, **F**: arrows). However, an extensive avascular area was observed in the left parietal lobe (round mark). SigS: sigmoid sinus; SS: straight sinus; TS: transverse sinus

microguidewire (Traxcess 14; Terumo, Tokyo, Japan) were guided to the occluded end of the left SigS. Coil embolization of the left SigS to TS was performed through this site. Although a small volume of shunt blood directly flowing in an area adjacent to the sinus confluence remained, antegrade outflow from the sinus confluence to the right TS was noted and reflux to the SS had disappeared; antegrade blood flow at the SS was observed (**Fig. 3**). At this point, we considered effective flow reduction to have been achieved and deep vein congestion to have been reduced. The operative time was 6 hours and 41 minutes. There were no remarkable changes in the blood pressure or vital signs during surgery. On final angiography, there was no extravascular leakage of contrast medium, but an extensive avascular area was observed in the left parietal lobe.

Postoperative course: Immediately after the treatment, brain CT was performed to confirm the presence of hemorrhagic complications. Extensive cerebral hemorrhage of the left parietal lobe was noted (**Fig. 4**). Hematoma removal under craniotomy was considered, but the patient's family did not wish for craniotomy; therefore, conservative treatment was selected and the patient died 31 days after surgery.

Discussion

Previous studies suggested that the risk factors for hemorrhage in patients with dAVF include symptomatic dAVFe, cortical venous reflux, outflow tract varices, posterior cranial fossa lesions, and advanced age. Cortical venous reflux may increase the venous pressure, causing cerebral hemorrhage. In particular, the localization of an outflow tract may lead to a rapid increase in the topical venous pressure, inducing hemorrhage. If morphological changes, such as varices, are present, the risk of hemorrhage may be higher.[1–6)]

On the other hand, some investigators emphasized that the risk of hemorrhage cannot be evaluated based on the presence of cortical venous reflux alone, which is described in Cognard's and Borden's classifications. Several studies reported that cortical venous reflux developed

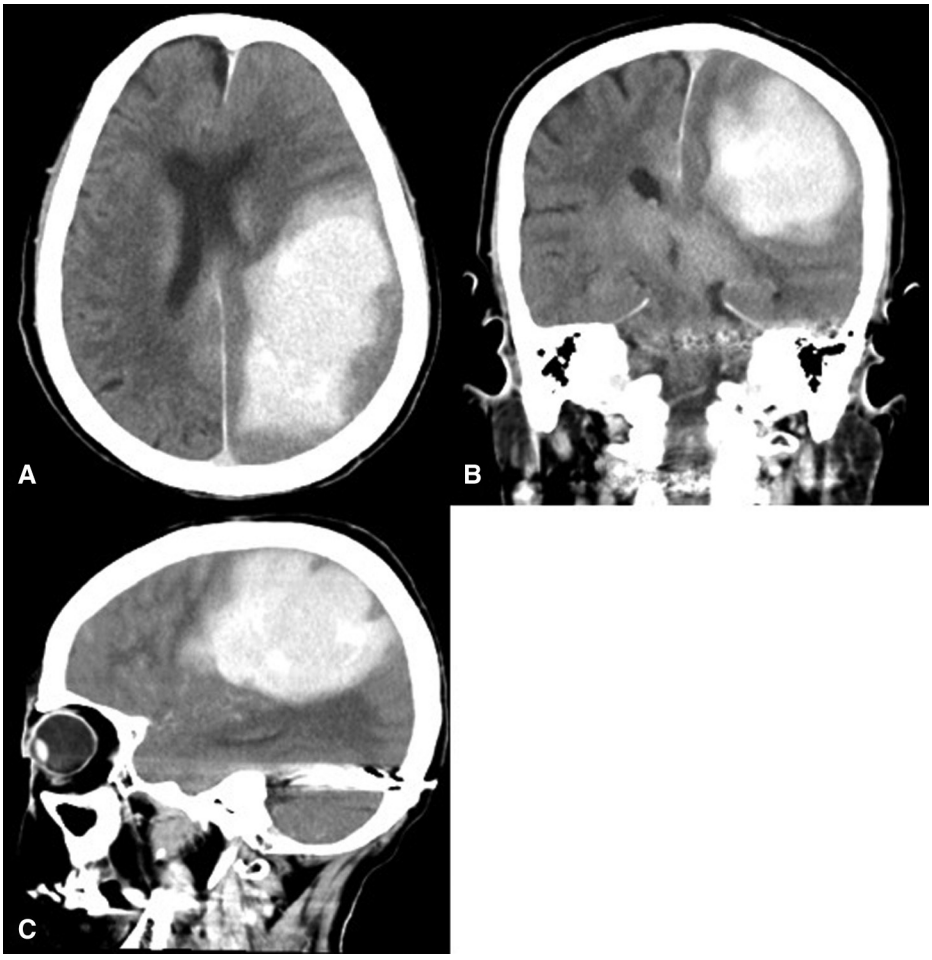

Fig. 4 (**A**) Axial section on postoperative CT. (**B**) Coronal section. (**C**) Sagittal section. Extensive cerebral hemorrhage involving the left parietal lobe was observed.

during follow-up in approximately 2% of Cognard type 1/2a or Borden type 1 patients, and that new shunts were noted in some cases.[7,8]

Stenosis or thrombosis of the sinus may also be involved in an increase in the venous pressure.[9] In particular, rapid changes in hemodynamics related to thrombosis of the sinus may lead to symptom changes or rapid progression, causing serious, irreversible changes in the brain such as intracerebral hemorrhage and venous infarction.

In the present case, an acute, symptomatic dAVF with cortical venous reflux was observed. Strom et al. and Gross et al. previously reported that the annual incidence of hemorrhage is approximately 10%–18.2%. In the present case, the timing of hemorrhage onset was unclear, but angiography demonstrated slight extravascular leakage of contrast medium; at this point, hemorrhage may have gradually begun. Hemorrhage may have developed a few hours after onset, being relatively early.[5,6]

Jolink et al. reported that the risks of death and unfavorable functional prognosis in patients with dAVF-related hemorrhage were lower than in those with hypertensive cerebral hemorrhage or rupture of arteriovenous malformation. As dAVF-related hemorrhage is caused by an increase in the venous pressure, the pressure may be lower than that in patients with hypertensive cerebral hemorrhage or rupture of arteriovenous malformation.[10] In comparison, the condition in the present case was rapid and serious.

Regarding the present case, we assumed the following hypothesis: A dAVF draining via the SS and left internal jugular vein corresponding to Cognard type 1/2a or Borden type 1 may become one draining via the SS only because of left internal jugular vein occlusion by thrombosis. Therefore, it may have induced rapidly progressing gait disorder and consciousness disturbance. This hypothesis is compatible with MRI findings and markedly rapid deterioration

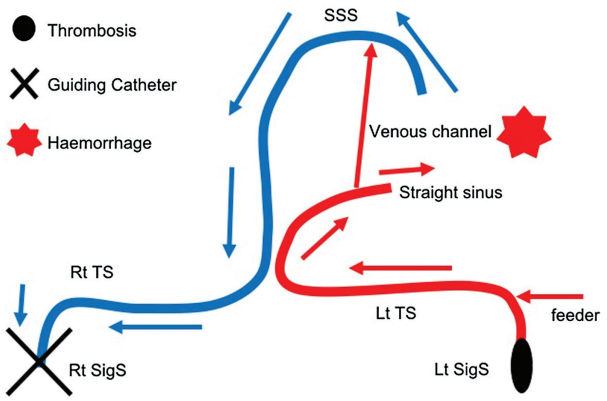

Fig. 5 The left internal jugular vein and sinus confluence were occluded. Shunt blood flow regurgitated in the deep and parietal lobe cortical veins, flowing into the SSS. The flow of SSS, right TS, SigS, and internal jugular vein was antegrade. The right internal jugular vein was the main outflow tract from the intracranial cavity, involving normal perfusion. However, a guiding catheter was inserted into this vein, which may have induced perfusion disorder. SigS: sigmoid sinus; SSS: superior sagittal sinus; TS: transverse sinus

(consciousness disorder 4 hours after onset). Brain MRI revealed edematous changes of the bilateral thalami; therefore, rapid deep vein congestion and an increase in the intracranial pressure may have progressed.

DSA demonstrated occlusion of the left SigS/TS/internal jugular vein. The main outflow tract from the intracranial cavity to the extracranial cavity was the right internal jugular vein alone. As other outflow tracts, only a few outflow routes from the venous plexus to the extracranial cavity remained. Simultaneously, slight extravascular leakage of contrast medium was observed. At this point, hemorrhage may have gradually begun. In addition, an wide-diameter guiding catheter (8 Fr) was inserted into the right internal jugular vein as a major outflow tract for transvenous embolization, and sufficient blood outflow from the SSS may have been hindered, inducing perfusion disorder of the bilateral cerebrums. This led to cerebral hemorrhage involving the left parietal lobe, in which cortical venous reflux was present, and marked venous congestion with leakage of contrast medium (**Fig. 5**).

First, to prevent rapidly progressing venous perfusion disorder, as demonstrated in the present case, transarterial embolization should be initially performed as a first-choice procedure to reduce the intraoperative venous pressure as much as possible through a decrease in blood inflow. Recently, effective embolic materials, such as Onyx, have become available, and if blood inflow had been reduced, we may prevent venous hypertension during transvenous embolization.

Second, if transvenous embolization had been performed through penetration of the left internal jugular vein, which may have been rapidly occluded, maintenance of the right internal jugular vein as the main outflow tract would have been possible, preventing the rapid increase in venous pressure. Furthermore, if a small-diameter (6 to 7 Fr) guiding catheter had been used to approach through the right internal jugular vein, the influence on venous return may have been reduced.

In the present case, consciousness disorder developed 4 hours after onset; rapid symptom deterioration was noted. Therefore, accurate reduction of shunt blood flow in a short time was prioritized. Initially, transarterial embolization was abandoned because type III aortic arch made access difficult. Furthermore, the left internal jugular vein was occluded and approaching was considered to be difficult. An approach through the right internal jugular vein, through which a route may be accurately secured, was selected. In addition, for coil embolization, it is necessary to penetrate the occluded sinus. To improve the supportiveness of a catheter, we used a thick-diameter (8 Fr) guiding catheter. This may have increased venous congestion, inducing intracranial hemorrhage.

Furthermore, to achieve hemostasis early after extravasation detection, we should have considered embolization of the posterior SS with reflux prior to TS/SigS embolization to initially block reflux. However, in this case, it was necessary to evaluate the collateral pathway of a deep vein.

Recently, endovascular therapy has been recognized as a first-choice treatment for dAVFe of the TS to SigS, but the drainage pattern must be accurately evaluated. In some cases, direct surgery should also be considered as an option.

Lastly, prompt, time-prioritized treatment is unavoidable for patients with rapid symptom progression, but it is important to evaluate the hemodynamics, particularly for drainage pattern thoroughly and establish accurate therapeutic strategies for the treatment of dAVFe. Physicians should be prepared for acute-phase treatment such that sufficient therapeutic strategies are made in a short time after calmly assessing the condition.

Conclusion

We report a patient on whom emergency transvenous coil embolization for an acute, symptomatic, non-hemorrhagic dAVF with cortical venous reflux was performed, but extensive cerebral hemorrhage developed immediately after surgery. In the treatment of dAVFe with rapid

symptom progression, prompt, time-prioritized treatment is unavoidable, but physicians should be prepared to accurately evaluate the condition in a short time to enable the prompt selection of satisfactory therapeutic strategies.

Disclosure Statement

We declare no conflict of interest.

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