Successful Stentings for Bilateral Jugular Bulb Stenosis/Occlusion in a Case of Bilateral Transverse Sinuses Dural Arteriovenous Fistulas: Case Report

Takahiro Murata, Hisashi Nagashima, Shunsuke Ichinose, Toshiya Uchiyama, Jun-ichi Koyama, and Kazuhiro Hongo

Objective: We describe successful stentings for treatment of bilateral jugular bulb stenosis and occlusion in a case of dural arteriovenous fistulas (d-AVFs) of bilateral transverse sinuses.

Case Presentations: A 71-year-old woman presented with rapidly progressive cognitive impairment as well as consciousness disturbance, and cerebral angiography demonstrated d-AVFs of bilateral transverse sinuses, right jugular bulb occlusion, and left jugular bulb stenosis accompanied by marked intracranial venous congestion. The symptoms were temporarily alleviated after transarterial and transvenous embolization, but recurred one year later. Curative embolization was thought to be difficult, and stenting for stenosis/occlusion of the bilateral jugular bulbs, which were considered to be causing her symptoms, was performed and resulted in improvements in the condition.

Conclusion: Stent for treating sinus stenosis/occlusion accompanying d-AVF is effective to improve venous congestion and hypertension.

Keywords ▶ dural arteriovenous fistula (d-AVF), jugular bulb, percutaneous transluminal sinus stenting, sinus thrombosis, sinus stenosis

Introduction

Dural arteriovenous fistula (d-AVF) is often accompanied by stenosis or occlusion of the venous sinus due to sinus thrombosis, and sinus stenosis/occlusion is closely involved in the pathological condition and progression of d-AVF. The etiology of d-AVF and sinus stenosis/occlusion is controversial. It is known to be developed secondary to head trauma, surgical craniotomy, or sinus thrombosis. Several factors, including thrombi in the sinus, venous hypertension, and inflammation have been reported as etiologic factors of d-AVF. On the other hand, the tendency that the presence of AV shunt promotes sinus thrombosis, leading to localization and resolution of the shunt with increases in the pressure in the sinus, has also been reported. Sinus stenosis/occlusion is one of the important factors in the treatment for d-AVF.

Here, we experienced successful stent placements of the bilateral jugular bulbs in a patient with stenosis and occlusion of the bilateral jugular bulbs accompanying d-AVF of the bilateral transverse sinuses (TSs), who had the onset with rapidly progressing dementia. Stenting resulted in reducing the increased venous pressure and succeeded in relieving the symptoms. We describe the details and discuss stenting for sinus stenosis/occlusion accompanying d-AVF.

Case Presentation

A 71-year-old woman developed an increased tendency to fall and cognitive impairment, 6 months before referral to us. She had a past history of pulmonary embolism and had taken oral warfarin. Her symptoms were progressive, and she became incapable of standing up or walking, and exhibited consciousness disturbance, 5 months after the onset. MR imaging demonstrated extensive edematous changes in the bilateral cerebral cortices, and MR angiography suggested d-AVF. On admission, consciousness disturbance was marked with the Glasgow Coma Scale (GCS) score varying from 6–11;
aphasia and tetraparesis were noted, and completely dependent in her activity of daily living (ADL) including nasal tube feeding. Cerebral angiography revealed a high flow d-AVF involving the confluence of sinuses to the left TS (Fig. 1). The fistulas were fed by multiple branches of the bilateral external carotid arteries, including the occipital artery (OA), the middle meningeal artery (MMA), muscular branches and posterior meningeal artery (PMA) from the vertebral artery (VA). The venous flow was primarily drained into the left jugular vein antegrade, but stenosis was observed in the bulb (Fig. 1). The right jugular vein was occluded in the bulb and drained into the vertebral venous plexus through narrow channels. There was retrograde venous flow in the superior sagittal sinus (SSS) and straight sinus with marked cortical venous reflux. On bilateral internal carotid arteriography, no antegrade outflow of the cerebral venous circulation toward the SSS was noted, and intracranial venous congestion was observed.

**Endovascular Therapy**

**1st intervention**

MR imaging showed minor intracerebral hemorrhage, semi-urgent treatment was judged to be required. The treatment consisted of maximally possible transvenous embolization (TVE) of the dilated parallel venous channels near the left TS using 23 coils (165.5 cm) and transarterial embolization (TAE) of the bilateral MMAs and OAs using a liquid embolic agent. Decreasing the shunt flow gradually resolved consciousness disturbance after surgery, the second embolization was scheduled for further improvements.

**2nd intervention**

TVE in the dilated parallel venous channels below the confluence of sinus was achieved using 22 coils (192 cm). With sufficient anticoagulation treatment, percutaneous transluminal angioplasty (PTA) was achieved using a PTA balloon (Gateway 4.0 × 20 mm, Stryker, Kalamazoo, MI, USA) at a low pressure (4atm) in the stenotic portion of the left jugular bulb. With alleviation of stenosis, the antegrade venous drainage increased towards the left jugular vein, and intracranial venous circulation improved comparably prior to surgery despite persistence of cortical venous reflux.

**Clinical course after the 1st and 2nd interventions**

The decrease in the shunt flow and alleviation of stenosis of the venous outflow tract resulted in disappearance of edema of the cerebral cortex on MR imaging. Neurologically, her GCS score improved to 14, and Mini-Mental State Examination (MMSE) to 10, she was able to take food orally and was ambulatory. After hospitalization for rehabilitation, she became independent 3 months after the 2nd intervention. However, gait instability and cognitive impairment gradually recurred 1 year after the 2nd intervention, and her consciousness deteriorated. An MR imaging showed extensive edematous changes in the bilateral cerebral cortices (Fig. 2), and cerebral angiography indicated
Stentings for Bilateral Jugular Bulb Stenosis/Occlusion with d-AVF

A 6-Fr guiding sheath (Shuttle, COOK MEDICAL, Bloomington, IN, USA) was placed in the right internal jugular vein, and the re-occluded right jugular bulb was recanallized using a microcatheter (Excelsior1018, Stryker) and a micro-guide wire (Chikai14, Asahi Intecc, Aichi, Japan). The micro-guide wire was exchanged to ChoICE PT Exchange 300 cm (Boston Scientific), and Aviator 4.0 mm × 20 mm (Cordis Endovascular, Miami Lakes, FL, USA) was used for pre-dilation, a self-expanding open-cell stent (Precise Pro RX 6.0 mm × 20 mm, Johnson & Johnson, Miami, FL, USA) was deployed to the occluded portion, and post-dilation was attempted using Shiden 5.0 mm × 20 mm (Kanaka Medix, Osaka, Japan) (Fig. 3). Next, a guiding sheath was advanced in the left jugular vein, the stenotic portion of the left jugular bulb was dilated as in the right jugular bulb using the same device, 2 stents (Precise Pro RX 8.0 mm × 30 mm) were deployed because of adjusting its curvature, and post-dilation was also attempted as the same manner (Fig. 3). Cerebral angiography after stent placements showed antegrade venous drainage of the cerebral circulation into the SSS, elimination of the cortical venous reflux from shunt, and the absence of acute occlusion or restenosis in the stents.

Results

Postoperative CT scan demonstrated that the stents were sufficiently expanded in the curved portion of the both jugular bulbs (Fig. 4). MR imaging after one month from stenting disclosed resolution of the cortical edema (Fig. 4), and MR venography clearly showed the patency of bilateral jugular veins (Fig. 4).
Consequently antiplatelet medication was reduced to one drug. Neurologically, the GCS score improved to 15, and the MMSE to 22. She was ambulatory and discharged to home independently in her ADL. Although d-AVF remained, she was followed up without any neurological deterioration in an outpatient clinic, and the bilateral internal jugular veins were patent on MR venography 9 months after stent placements.

Discussion

In endovascular treatment for d-AVF, curative treatment is generally attempted by transvenous embolization in the affected venous sinus using coils. However, patients with diffuse AV shunts in the sinus and those with a shunt at the confluence of sinuses are difficult to treat, because occlusion of the affected sinus may cause serious impairment of cerebral venous drainage. The case presented here also had refractory d-AVF primarily involving the confluence of sinuses, and treatment decision was made to improve the intracranial venous drainage by relieving venous outflow tract obstruction because she showed marked neurological deterioration. In this case, d-AVF, which was preoperatively Borden Type II, became Type I after stent placement, and her clinical symptoms were markedly alleviated. This
suggests that an increase in the venous pressure by two mechanisms, i.e., influx of arterial blood into the venous sinus via the shunt and outflow tract obstruction due to stenosis or occlusion of the venous sinus, is closely involved in the pathology of d-AVF. There have been a few reports of treatment in patients with stenosis/occlusion of the venous sinus associated with d-AVF for relieving venous outflow tract obstruction. Haraguchi et al. presented a case of transverse sinus-sigmoid sinus (TS-SS) d-AVF in which PTA achieved for marked stenosis of the contralateral TS-SS prior to radical treatment was useful and reported that, by measuring the pressure in the sinus before and after PTA, improvement of intracranial venous congestion and decrease in the pressure in the sinus could be confirmed after PTA. In the reported case, treatment was PTA alone without stent placement. In our case, stent was not placed at the 2nd intervention, because intracranial venous congestion was alleviated, and antegrade venous outflow was improved by dilation of the stenotic portion; however, stenting was eventually needed due to restenosis.
Stenting of the venous sinus for the treatment of d-AVF has been reported with recent advancements in the interventional techniques and devices.\textsuperscript{5–11} In most reported cases, stenosis/occlusion was present in the affected sinus with AV shunt, and stent was placed in expectation of obliteration of the shunt site by compressing the sinus wall as well as preservation of the antegrade venous outflow tract by dilating the sinus. On the other hand, in a small number of reported cases, stents have been placed in the stenotic/occluded portion of the venous sinus distant from the affected sinus similar our case.\textsuperscript{7} Stents have often been placed not only in the TS to SS,\textsuperscript{6–8} but also in the SSS\textsuperscript{9,10} or straight sinus.\textsuperscript{5,11} The stents used were often balloon- or self-expanding closed-cell type,\textsuperscript{5–7,9,11} but a few instances of balloon-expanding covered stent have also been reported.\textsuperscript{8,10} There have been no reports on stenting for stenosis/occlusion of the jugular bulb and placement of an open-cell stent in the sinus in the literatures. Since the jugular bulb is surrounded by the skull base and markedly curved, we obtained favorable results by deploying open-cell stents with excellent conformation to curvature.

However, as stents were placed in the venous sinus while persisting d-AVF in our case, prevention of restenosis/reocclusion due to in-stent thrombosis is an important issue. Levrier et al.\textsuperscript{7} reported stent placement in ten patients with the TS-SS d-AVF and observed postoperative cerebral hemorrhage in one patient for whom postoperative antithrombotic treatment was not given. This hemorrhage was suspected to be caused by occlusion of the cortical vein due to thrombosis, they administered clopidogrel from before surgery until 3 months after surgery in the subsequent patients. Spiotta et al.\textsuperscript{10} reported a case that received placement of a covered stent for d-AVF of the SSS to the confluence of sinuses but developed occlusion of the SSS due to in-stent thrombosis on the 4th postoperative day despite sufficient postoperative antithrombotic treatment consisting of oral administration of aspirin and clopidogrel and intravenous administration of heparin. This reported patient had a factor V Leiden thrombophilia liable to venous thromboembolism, which suggested inducing in-stent thrombus formation. Since our patient also had a past history of pulmonary embolism, she might have some latent clotting abnormality. As she exhibited a slight sign of cerebral hemorrhage at the initial intervention, the shunt flow was reduced by TVE and TAE, and PTA was achieved at the 2nd and 3rd interventions with sufficient anticoagulation therapy. Currently, there is no consensus as to perioperative antithrombotic therapy or its period for stenting in the venous sinus, and they vary with the type of stent. However, there have been many reports of oral antiplatelet medication combined with intravenous or oral administration of anticoagulants in the acute period,\textsuperscript{8,10,11} and the duration of antiplatelet medication was diverse. At the 4th intervention, we placed stent grafts with sufficient antithrombotic therapy from before surgery using dual antiplatelets and an anticoagulant and could obtain a favorable outcome without occlusion. d-AVF is closely related to venous sinus thrombosis, and since there may be hypercoagulable state or nonhereditary thrombogenic tendency such as protein C deficiency in the patient’s background, indications for stenting in patients with d-AVF must be carefully evaluated. However, satisfactory results may be obtained by stepwise treatment from PTA to stenting and appropriate antithrombotic therapy as well as proper selection of patients.

\section*{Conclusion}
A case of bilateral TSs d-AVF successfully treated by stenting for stenosis and occlusion of the bilateral jugular bulbs was presented. Stent placement is effective for the treatment of stenosis/occlusion of the venous sinus causing venous outflow tract obstruction or venous hypertension. d-AVF is closely related to venous sinus thrombosis, and stenting for such conditions requires antithrombotic treatment appropriate for each case.

\section*{Disclosure Statement}
None of the authors has conflicts of interest in reporting the present case.

\section*{References}


