Dural arteriovenous fistulas (DAVs) are relatively uncommon (incidence of 0.15–0.16 cases/100,000 people/year) and represent approximately 10%–15% of all intracranial arteriovenous shunts. The clinical course of DAVF is variable, ranging from asymptomatic to symptomatic presentations, with an aggressive presentation consisting of venous hypertension and neurological deficit in patients with high-flow shunts. In patients with cortical venous reflux, intracerebral hemorrhage (ICH) may be the initial symptom. The presence of deep venous drainage causes venous infarction, resulting in ICH and dementia. Most DAVFs are treated via an endovascular approach; however, successful obliteration of complex DAVFs may require a combined approach involving open surgery, endovascular treatment, and radiosurgery. Routine endovascular techniques include transarterial and transvenous embolization by coil placement and injection of particles or liquid embolic material as an adjunct to surgical treatment or as a curative treatment. In some patients, however, the use of these techniques is impossible given the associated problems. Here we describe the details in a case of torcular DAVF in which the complete obliteration of shunt flow was achieved by stent placement and angioplasty. We describe the technical details of this case.

Case Report

History and Examination

A 64-year-old man with progressive dementia (Mini-Mental State Examination [MMSE] Score 13) was referred to our department with the diagnosis of torcular DAVF. Six years earlier, he had undergone transarterial and transvenous embolization for a cavernous DAVF that presented with headache, conjunctival injection, and exophthalmos; complete obliteration of the fistula orifice with self-expandable stents and angioplasty (balloon inflation) is considered the best treatment option to preserve normal cerebral venous sinus drainage and obliterate shunt flow. In such cases, the authors recommend using one or more self-expandable and closed-cell stents and using angioplasty to avoid endoleakage into the gap between the stent graft and the vessel wall.


KEY WORDS torcular dural arteriovenous fistula; stent placement; angioplasty; vascular disorders
room with left paresis and sensory disturbance. Computed tomography scanning on admission demonstrated a right thalamic hemorrhage (Fig. 1). Cerebral angiography revealed a torcular DA VF fed by multiple meningeal arteries with cortical and deep venous reflux and venous congestion (Borden Type II). Transarterial embolization was performed several times, and open surgery (sinus isolation) was performed once. Although flow in the DA VF decreased after these treatments, complete obliteration was not achieved. In addition, the multiple treatments administered caused skin flap ulceration. Thus, the skin was reconstructed with a latissimus dorsi musculocutaneous flap (anastomosis: the superficial temporal artery with the dorsal thoracic artery, the superficial temporal vein with the dorsal thoracic vein). After these therapies, cognitive function improved and venous reflux decreased. However, during the follow-up period, dementia worsened over time. We performed FLAIR MRI, which showed hyperintensities in bilateral thalami, indicating cerebral edema (Fig. 2A and B). In addition, T2*-weighted images showed hypointensities in bilateral thalami, which indicated previous hemorrhage and microbleeds (Fig. 2C and D).

Angiographic Findings

Digital subtraction angiography demonstrated multiple arteries feeding the torcular DA VF from the intracranial and extracranial circulations. Cerebral angiography revealed increased shunt flow along with cortical and deep venous reflux. Contrast injection in the right internal carotid artery (ICA) revealed filling of the DA VF from the tentorial arteries and posterior cerebral arteries. Contrast injection in the left ICA revealed filling of the DA VF from the tentorial arteries. Contrast injection in the right external carotid artery (ECA) demonstrated filling of the DA VF from the superficial temporal arteries and occipital arteries. Contrast injection in the left ECA demonstrated filling of the DA VF from the middle meningeal arteries and superficial temporal arteries. Contrast injection in the right vertebral artery (VA) revealed filling of the DA VF from the posterior cerebral arteries. Antegrade venous drainage was directed toward the right transverse sinus, and retrograde venous drainage was directed toward the deep venous system through the straight sinus. Markedly dilated cortical and deep veins and right transverse sinus stenosis were observed (Fig. 3).

Operation

The patient and his family provided written informed consent for the procedure, which was performed under general anesthesia. The right femoral artery was accessed using a 5-Fr sheath, and a 4-Fr diagnostic catheter was placed in the left ECA to guide contrast injections. The right jugular vein was accessed with an 8-Fr sheath, and an 8-Fr guiding catheter (Brite Tip straight guiding catheter, Cordis, Johnson & Johnson) was introduced over a hydrophilic 0.035-in soft-tip angled guidewire and was navigated into the right transverse-sigmoid junction. A percutaneous transluminal angioplasty balloon (3.5 × 20 mm, Ryujin Plus OTW, Terumo) was dilated at the right stenotic portion of the right transverse sinus, and a self-expanding closed stent (8 × 29 mm, Carotid Wallstent Monorail, Boston Scientific Ireland Ltd.) was deployed from the straight sinus to the right transverse sinus, covering the stenotic portion of the right transverse sinus and the affected straight sinus. Contrast injection in the left ECA still demonstrated filling of the DA VF. Therefore, to deploy another self-expanding closed stent over the first stent, we attempted to introduce it into the right transverse sinus. Because it was difficult to advance it into the right sinus, another stent was deployed. Contrast injection in the left ECA was repeated after the procedure, showing no filling of the DA VF.
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transverse sinus, we replaced the straight guiding catheter with an angled guiding catheter (8-Fr angled peripheral guide catheter, Launcher, Medtronic). We introduced the angled guiding catheter and the second stent (8 × 21 mm, Carotid Wallstent Monorail) over the wire into the first stent, pulled the guiding catheter, and deployed the second stent over the first stent in the affected sinus. Contrast injection in the left ECA still demonstrated filling of the DAVF, but shunt flow was markedly decreased. Contrast injection in the right ECA demonstrated filling of the DAVF; therefore, we deployed a third stent (8 × 20 mm, Iliac Wallstent RP, Boston Scientific Ireland Ltd.) over the first and second stents at the lesion of the shunt point in the same manner. After successful deployment of the stent graft, repeat balloon inflations (4.5 × 20 mm, Sterling Balloon Dilation Monorail Catheter, Boston Scientific USA) were performed to fix the stent graft to the venous sinus wall. Contrast injection in the bilateral ECA, ICA, and VA still demonstrated filling of the DAVF, but retrograde venous drainage was absent (Borden Type I; Fig. 4).

FIG. 3. Bilateral ECA, bilateral ICA, and right VA angiograms demonstrating a torcular DAVF. Right ECA angiograms demonstrated the DAVF feeding of the right superficial temporal artery and right occipital artery in the anteroposterior projection (early arterial phase, A) and lateral projection (early arterial phase, B). Left ECA angiograms demonstrated filling of the DAVF from the left middle meningeal artery and left superficial temporal artery in the anteroposterior projection (late arterial phase, C). Right ICA angiograms demonstrated filling of the DAVF from the right tentorial artery and right posterior cerebral artery in the anteroposterior projection (late arterial phase, D) and lateral projection (early arterial phase, E). Left ICA angiograms demonstrated filling of the DAVF from the left tentorial artery in the anteroposterior projection (late arterial phase, F). Right VA angiograms demonstrated the DAVF fed by the right posterior cerebral artery in the anteroposterior projection (late arterial phase, G). Antegrade venous drainage was directed toward the right transverse sinus, and retrograde venous drainage was directed toward the deep venous system through the straight sinus. Right VA angiograms demonstrated markedly dilated vein of Galen, internal occipital vein, and internal cerebral vein in the lateral projection (venous phase, H), and left ECA angiograms demonstrated right transverse sinus stenosis in the anteroposterior projection (late arterial phase, I). Left ECA 3D angiograms demonstrated a torcular DAVF feeding of the left middle meningeal artery and left superficial temporal artery (I).
Postoperative Course

The patient was discharged on the 4th postoperative day and was placed on 200 mg of cilostazol and 15 mg of rivaroxaban daily. At the 3-month follow-up, the patient's neurological findings had ameliorated (MMSE Score 23). Follow-up angiography performed at that time demonstrated normal cerebral venous drainage with in-stent stenosis of the straight and right transverse sinuses and without recurrent shunt flow (Fig. 5A–D). The hyperintensity in bilateral thalami observed on FLAIR images had decreased (Fig. 5E and F).

Discussion

Abnormal arteriovenous communication located in the sinus wall is generally considered to be the pathology in DAVFs; however, their pathogenesis is not fully understood. A limited number of histological evaluations of resected specimens have shown a constant pattern in the architecture of the affected sinus wall. The essential abnormality was not a direct arteriovenous shunt to the sinus lumen; rather, it was a connection between dural arteries and dural veins within the venous sinus wall.4,11

Dural AVFs are addressed with a multimodal approach involving endovascular treatment (transarterial and transvenous embolization), open surgery, and radiosurgery. However, these approaches were not applicable in the present case for the following reasons. 1) During transvenous embolization of DAVFs, in general, the venous pouches, parasinuses, and affected sinuses are emboлизed using detachable coils; however, it is important to preserve normal venous drainage. In the present case, the affected sinus was the straight sinus and the right transverse sinus near the torcular herophili. We wanted to accomplish complete obliteration of the DAVF and preserve normal venous drainage from the straight sinus to the right transverse sinus. 2) During transarterial embolization using detachable coils and liquid embolic agents, if the shunt points are embolized, complete obliteration of the DAVF can be accomplished; however, this is very difficult. Dural AVFs occasionally recur because their feeding arteries have extensive collateral networks. In the present case, the skin was reconstructed using a latissimus dorsi musculocutaneous flap. Transarterial embolization can result in avascularity of the skin and skin ulceration; thus, the procedure was not applicable in the present case. 3) Surgical isolation or resection of the fistulous sinus is associated with a high cure rate; however, it is not always feasible and carries the risks associ-
ated with open surgery, such as blood loss, brain adhesions, and skin flap problems. In the present case, the multiple treatments administered caused skin flap ulceration, and the skin was reconstructed with a latissimus dorsi musculocutaneous flap. We thought that open surgery would certainly result in skin flap troubles; therefore, we did not consider open surgery in the present case. 4) Recent studies on the use of stereotactic radiation therapy reported a relatively high occlusion rate (in approximately 60% of cases) several months after treatment, without significant complications; however, DAVF obliteration via radiosurgery is a long process. We needed to obliterate the shunt flow and retrograde venous drainage as soon as possible. Therefore, we did not consider radiosurgery in the present case.

If stent placement in the affected sinus can obliterate a DAVF shunt, it can decrease shunt flow immediately and preserve the straight and right transverse sinuses. Choi et al.\textsuperscript{3} reported the use of a stent graft to preserve venous sinus flow in a DAVF involving the dominant transverse-sigmoid junction and sigmoid sinus in a patient with hypoplasia of the contralateral venous sinuses and intolerable balloon occlusion test for the ipsilateral venous sinuses. Stent placement in the affected sinus of DAVFs and angioplasty could compress these vessels in the sinus wall and occlude the arteriovenous shunt flow hemodynamically. Therefore, in cases in which there is no evident shunt point, such as the venous pouches and parasinuses, but there is diffuse shunt flow in the affected sinus, stent placement with or without angioplasty in the affected sinus can be an effective treatment to occlude the DAVF and preserve normal cerebral venous sinus drainage.\textsuperscript{1,2,4,6–9,12,15}

Stent selection among patients with DAVF is a matter of some debate. An investigation performed by Levrier et al.\textsuperscript{6} showed that the inner endoluminal compression force on the sinus wall is related to the stent radial force and is dependent on the mechanical characteristics of the stent. Stainless steel stents usually provide a stronger radial force than nitinol stents. These authors reported that stent caliber size should be appropriate. An undersized stent will not compress the sinus wall and will not significantly decrease shunt flow. They also asserted that stent lengths should be appropriate. Insufficient covering of the affected sinus leads to incomplete treatment. A study performed by Ohara et al.\textsuperscript{12} indicated that self-expandable and closed-cell type stents, such as the Wallstent, are suitable for the recanalization of thrombosed sinus in DAVFs because their radial force may also induce compression of the fistulous dural wall of the affected sinus. Choi et al.\textsuperscript{3} and Liebig et al.\textsuperscript{7} revealed that it is important to avoid blood leakage (endoleakage) into the gap between the stent graft and the vessel wall by repeated and high-pressured balloon inflation. The use of covered stents to treat a DAVF involving the transverse sinus has also been reported.\textsuperscript{3} Covered stents are effective in occluding perpendicular shunt flow to the stent strut. In the present case, we first selected a self-expandable closed-cell stent, the Wallstent, and used 3 closed-cell stents and performed repeated angioplasty to compress them on the affected sinus wall. We did this because closed-cell stents have a smaller free-cell area than open-cell stents; thus, an increase in the number of closed-cell stents yielded a smaller free-cell area. We chose multiple stent placements and angioplasty rather than a covered stent or a flow diverter because it is difficult to advance a covered stent into the right transverse sinus.
and straight sinus and because a flow diverter has still not been approved in Japan.

We attained a marked decrease in shunt flow and significant amelioration of clinical symptoms in the present case.

Another important concern during stent placement in the venous sinus is antithrombotic therapy. There is controversy regarding the selection and duration of antithrombotic agents. Choi et al. described a patient with a DAVF of the transverse sigmoid sinus treated via the placement of a balloon-expandable and covered stent graft in the affected venous sinus. In that case, premedication was not administered because the patient underwent emergenc treatment. After the procedure, the patient received 100 mg of aspirin and 75 mg of clopidogrel daily. In addition, low-molecular-weight nadroparin calcium (2850 IU/0.3 ml) was subcutaneously administered twice a day for 3 days. Follow-up angiography performed 9 months after the procedure demonstrated normal cerebral venous drainage without in-stent stenosis of the transverse-sigmoid junction. Spiotta et al. described a patient with a DAVF involving the superior sagittal sinus and torcular herophili who was treated with covered stent graft placement in the affected sinus. In that case, after the procedure, the patient received aspirin and clopidogrel daily, and heparin was intravenously administered. On postoperative Day 4, however, the patient became acutely unresponsive, developed uncal herniation syndrome, and died because of in-stent superior sagittal sinus occlusion. A study conducted by Levrier et al. demonstrated that stent graft placement can induce the coagulation process and may lead to cortical vein occlusion if appropriate anticoagulation therapy is not administered during the time required for epithelialization of the stent by endothelial cells. Thus, those authors recommend the use of antiplatelet agents for 3 months after stent placement in the cerebral venous sinus. In our case, after the procedure, the patient received 200 mg of cilostazol and 15 mg of rivaroxaban daily, and 3-month follow-up angiography demonstrated normal cerebral venous drainage without in-stent occlusion. Although the piling of several stents in the affected sinus of the DAVF was effective in the present case, we believe that appropriate antithrombotic therapy, such as a combination therapy of antiplatelet and anticoagulation agents, is also important. However, the duration of antithrombotic therapy is controversial. The patient will take 200 mg of cilostazol and 15 mg of rivaroxaban daily throughout his life. It is essential that follow-up cerebral angiography or CT venography is performed.

References


Author Contributions

Conception and design: Takada. Drafting the article: Takada. Critically revising the article: Takada, Isaka. Administrative/technical/material support: Takada, Isaka, Nakakuki, Mitsuno. Study supervision: Kaneko.

Correspondence

Shigeki Takada, Department of Neurosurgery, Hikone Municipal Hospital, 1882 Hassaka-cho, Hikone-shi, Shiga-ken 522-8539, Japan. Email: tshigekidesu@gmail.com.