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Case Report

The Case of a Patient with Idiopathic Intracranial Hypertension Who Required Additional Stenting for Stent-Adjacent Stenosis

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Objective: We report the case of a patient with recurred idiopathic intracranial hypertension (IIH) with transverse sinus (TS) stenosis after initial stenting, which was treated with additional stent placed in tandem to the secondarily occurred stent-adjacent stenosis (SAS).

Case Presentation: A 41-year-old woman complained of reduced visual acuity and blurred vision, and presented with papilledema. Lumbar puncture revealed an opening pressure of 36 cmH_2O. MRI revealed no space-occupying lesions, and the patient was diagnosed with IIH based on the modified Dandy criteria. MR venography revealed stenosis in the right and hypoplastic left TS. The patient complained of headache and neck pain after each lumbar puncture for examination. Venous sinus stenting (VSS) was performed in the right TS. One month after stenting, follow-up angiography revealed stenosis in the remaining parts of TS. Five months after stenting, IIH recurred, and SAS was detected on angiography. An additional stenting procedure was performed. Three months after the second treatment, her symptoms disappeared and cerebrospinal fluid pressure was normalized.

Conclusion: Patients with post-VSS recurrent IIH may develop restenosis in the remaining parts of TS at variable progression speeds. In this case, angiography revealed gradually advancing stenosis that seemed to form SAS at the time of recurrence. If the initial VSS is effective for IIH, SAS can also be treated effectively and less invasively with a second stent placement covering the entire TS length.

Keywords ▶ idiopathic intracranial hypertension, venous sinus stenting, stent-adjacent stenosis

Introduction

Idiopathic intracranial hypertension (IIH), also called pseudotumor cerebri and benign intracranial hypertension, is a condition that is now generally diagnosed as per the modified Dandy criteria[1] and manifests as elevated intracranial pressure (ICP) without any space-occupying lesion in the intracranial space, brain edema, or obvious increased cerebrospinal fluid. Papilledema is caused by IIH in 95% of the cases and may cause progressive optic nerve atrophy and visual disturbance, leading to blindness in 1%–2% of the patients.[2,3] Dural sinus stenosis exists commonly in IIH, and stent placement for the stenotic site is effective in patients who cannot be treated medically.[4] However, in some patients, recurrence is often caused by stent-adjacent stenosis (SAS), and the etiology for this is unknown.[5] We report the case of a patient with IIH accompanied by transverse sinus (TS) stenosis that recurred after the first stenting for the stenotic site as SAS, which was treated with tandem placement of additional stenting resulting in the normalization of intracranial hypertension.

Case Presentation

A 41-year-old woman had been experiencing headache, visual disturbance in the right eye, and blurred vision in both the eyes for several months and consulted an ophthalmologist. Her eyesight of the right and left eyes was 0.06 (corrected visual acuity [CVA]: 1.0) and 0.07 (CVA: 1.0), respectively. Papilledema was observed on the

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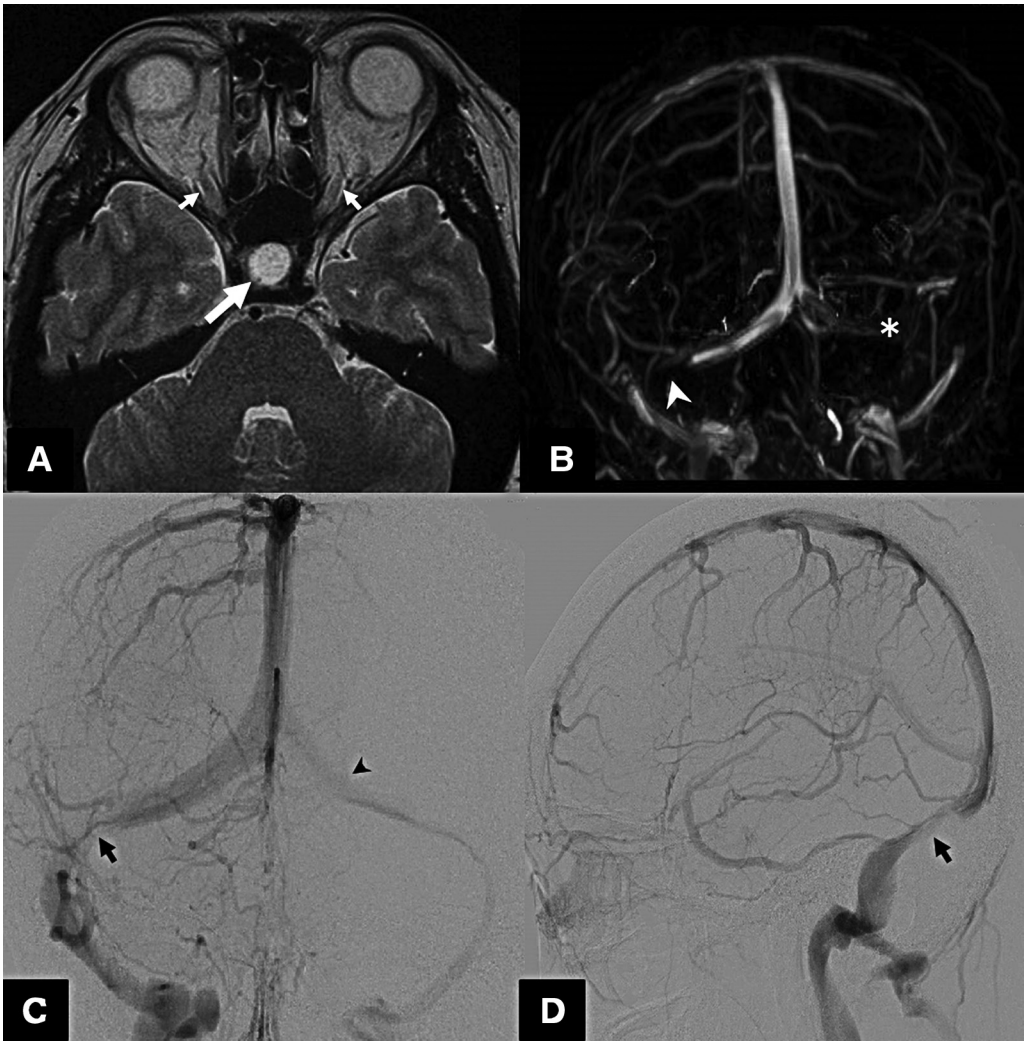

Fig. 1 (**A**) MRI revealing empty sella (large arrow) and dilated perineural subarachnoid space of the optic nerves (small arrows). (**B**) MRV shows stenosis of the right TS (arrowhead) and the left hypoplastic sinus (asterisk). (**C** and **D**) Frontal (**C**) and lateral (**D**) views of the right internal carotid angiogram reveal stenosis of the lateral portion of the right TS (arrows) and the hypoplastic left TS (arrowhead). MRV: MR venography; TS: transverse sinus

bilateral fundus. She was referred to our hospital for further examination.

Physical examination revealed obesity, with a body mass index of 28.8 kg/m^2. Neurological examination revealed no deficits. MRI showed empty sella and enlarged subarachnoid space around the optic nerves (**Fig. 1A**); MR venography (MRV) showed stenosis in the right TS and hypoplastic left TS (**Fig. 1B**). Lumbar puncture revealed an opening pressure of 36 cmH_2O, and the cerebrospinal fluid examination showed normal results.

She was started on a weight-loss program and acetazolamide (250 mg/day) medication. There was no improvement with respect to visual acuity and papilledema after 3 months. We planned lumboperitoneal shunt; however, she complained of headache and neck pain after lumbar puncture for examination. Therefore, we changed the treatment strategy from surgery to intervention. Cerebral angiography showed stenosis in the right TS and hypoplastic left TS (**Fig. 1C** and **1D**). The diameter of the narrowest portion of the right TS was 1.3 mm and that of the medial portion of the right TS and the right sigmoid sinus were 6.5 mm and 7.5 mm, respectively. Under local anesthesia, intravenous pressure was measured using an Excelsior XT-27 microcatheter (Stryker, Kalamazoo, MI, USA), which was navigated to the superior sagittal sinus (SSS) using a CHIKAI 300-cm microguidewire (0.014 inch; Asahi Intecc, Aichi, Japan). The venous pressure was 50 mmHg on the medial side of the right TS and 27 mmHg on the lateral side; the pressure gradient was 23 mmHg. Six months thereafter, we performed endovascular stenting

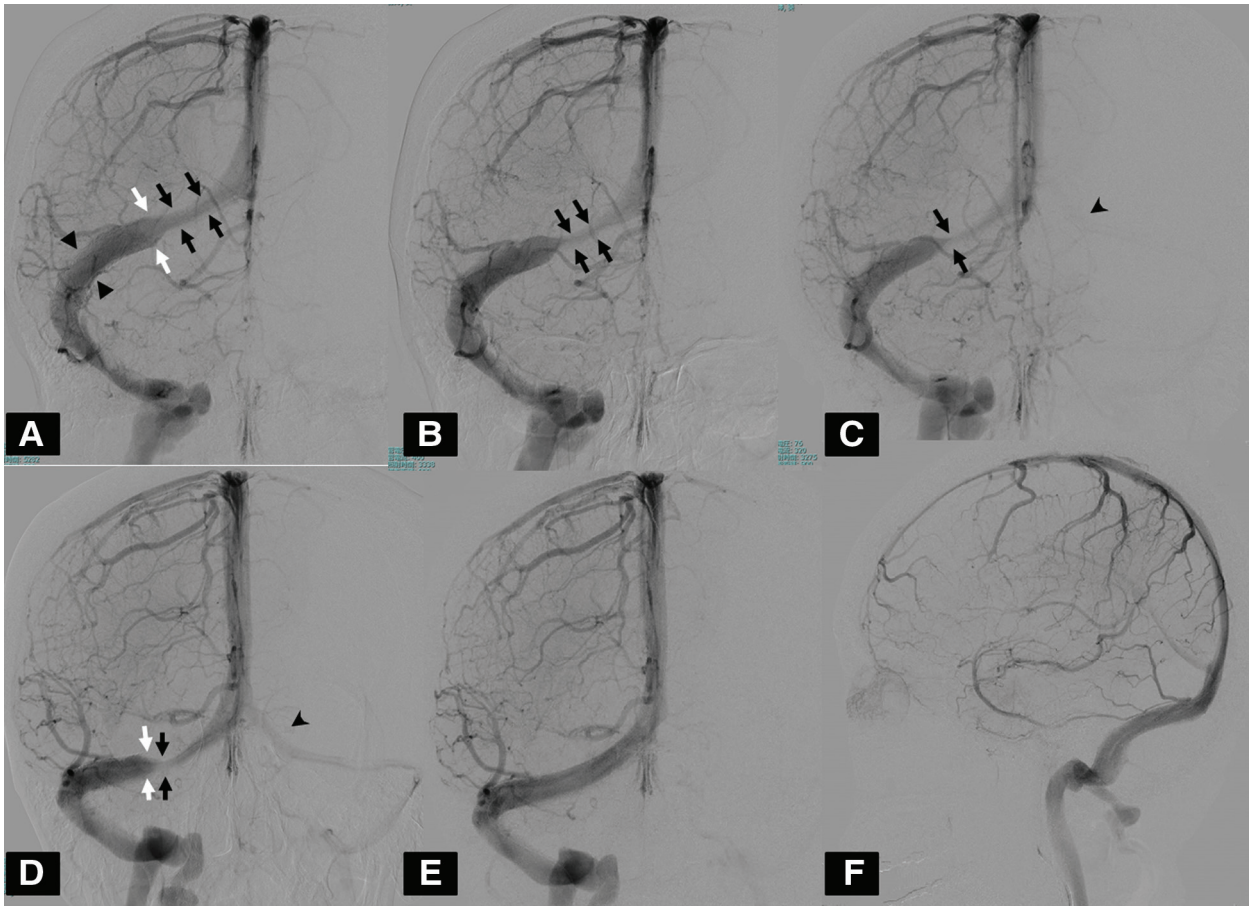

Fig. 2 DSA after initial treatment reveals dilated sinus (arrowheads) with stent on the day of stenting and slight stenosis (black arrows) (**A**) and progressing stenosis (black arrows) in the residual TS medial to the edge of the stent (white arrows) at 1 month (**B**) and 5 months (**C**) after stenting. Diagnostic DSA on the day of second stenting reveals SAS with an indistinct contrast medium at the narrowest portion near the medial edge of the initial stent (white arrows) (**D**). Blood flow draining the right TS (arrowheads in **C** and **D**) is also depicted. (**E** and **F**) DSA after second stenting shows dilated SAS with the second stent. DSA: digital subtraction angiography; SAS: stent-adjacent stenosis; TS: transverse sinus

with the approval of the ethics committee of our hospital (approval number 019006K).

First endovascular treatment

The first endovascular treatment that comprised dual antiplatelet therapy (DAPT) using aspirin (100 mg/day) and clopidogrel (75 mg/day) was started 7 days before the procedure. Under general anesthesia (GA), a 4-Fr catheter (JB2, Medikit, Tokyo, Japan) was inserted into the right femoral artery and placed in the internal carotid artery for angiography during the procedure. A 7-Fr sheath introducer was inserted into the right femoral vein and an intravenous heparin bolus (100 U/kg) was administered. A 7-Fr Launcher (Medtronic, Minneapolis, MN, USA) guiding catheter was placed in the right jugular vein at the level of the first cervical vertebra using a coaxial system of a 5F 125-cm JB2 catheter (Medikit, Tokyo, Japan) and a 0.035-inch 180-cm Radifocus guidewire (Terumo, Tokyo, Japan). We measured the intravenous pressure at a point medial to the lateral side of the stenosis using Excelsior XT-27 navigated using a CHIKAI microguidewire. The venous pressure on the medial and lateral sides of the right TS was 14 and 7 mmHg, respectively; the pressure gradient was 7 mmHg. After predilatation using a Sterling 3.5 × 30 mm (Boston Scientific, Marlborough, MA, USA), a PRECISE Pro RX stent 8 × 40 mm (Cordis, Miami, FL, USA) was placed at the stenotic site; this was done while ensuring that the midpoint of the stent was located at the narrowest point of the dural sinus stenosis. The vein of Labbe was involved within the stent. Angiography revealed sufficient dilation at the stenotic site (**Fig. 2A**). Postdilatation was not assessed. After stenting, the venous pressure on the medial and lateral side of the right TS was 11 and 9 mmHg, respectively; the pressure gradient was 2 mmHg. The pressure gradient in the venous sinus decreased from 7 to 2 mmHg after stenting. No complications occurred. For 4 weeks, DAPT was continued, and for the subsequent 8 weeks, clopidogrel was administered.

Four weeks after the stenting, follow-up angiography was performed under local anesthesia. The angiography findings included tapering of the right TS toward the lateral side—the narrow region of which has a certain length and is not restricted to a point—that was not typical SAS (**Fig. 2B**). The venous pressure was 22 mmHg on the lateral side of the right TS and 15 mmHg in the stent; the pressure gradient was 7 mmHg. The flow in the vein of Labbe was intact, and acetazolamide administration was ceased at this time. The cerebrospinal fluid pressure on lumbar puncture was reduced to 22 cmH_2O 6 weeks after stenting.

Her headache disappeared, and the visual acuity of right and left eyes was 0.05 (CVA: 1.2) and 0.08 (CVA: 1.2), respectively. Papilledema in the right eye reduced, whereas that in the left did not change.

Five months after stenting, she experienced a recurrence of headache. The results of her ophthalmologic examination remained unchanged. Angiography revealed progression of the TS stenosis and a prominent diameter gap at the site just medial to the stent. Furthermore, the contralateral TS was slightly visible (**Fig. 2C**). The venous pressure was 22 mmHg on the medial side of the right TS and 6 mmHg in the stent. The pressure gradient rose to 16 mmHg. The cerebrospinal fluid pressure on lumbar puncture was 30.5 cmH_2O.

Seven months after the first stenting, we performed the second intervention.

Second endovascular treatment

After 7 days of DAPT, we performed the second endovascular treatment under GA. Cerebral angiography revealed SAS and an indistinct contrast medium at its narrowest portion (**Fig. 2D**). The venous pressure on the medial and lateral sides of the right TS was 10 and 5 mmHg, respectively; the pressure gradient was 5 mmHg. The straight sinus entered via the right TS at a distance from the confluence; the diameter of this part of the TS was 6.8 mm. We placed a PRECISE Pro RX stent 7 × 30 mm from the point just lateral to the straight sinus entry point to the stent that was placed the last time, including the site of SAS. The two stents slightly overlapped; however, the diameter gap and contrast retention were not observed on angiography. Stenosis was released, and no perioperative complications were observed (**Fig. 2E** and **2F**). The venous pressure was 7 and 6 mmHg on the medial side of the right TS and in the stent, respectively; the pressure gradient was 1 mmHg. The venous pressure gradient was 5 mmHg before the second stenting and was reduced to 1 mmHg after stenting.

Her headache disappeared, and the cerebrospinal fluid pressure on lumbar puncture was decreased to 12.5 cmH_2O at 12 weeks after the second stenting.

Three months thereafter, visual acuity of the right and left eyes was 0.08 (1.2) and 0.08 (1.2), respectively. Papilledema remained unchanged. MRI showed disappearance of the finding of empty sella (**Fig. 3**).

Discussion

It has been clarified that endovascular venous sinus stenting (VSS) is more effective and safe than other treatments, including cerebrospinal fluid (CSF) diversion and optic nerve sheath fenestration (ONSF) for IIH accompanied by cerebral venous sinus stenosis. The improvement rate for headache and papilledema that received VSS was 77% and 93%, respectively. The effective ratio was 55% and 70% with CSF diversion and 26% and 92% with ONSF.[4] However, the risk of stenting is low. The prevalence of major complications, such as brain hemorrhage, venous thrombosis with secondary brain ischemia, and retroperitoneal hematoma was <2%.[5]

It is considered that stenting is effective when the pressure gradient across the stenotic site is high. Therefore, pressure gradient >10 mmHg is an indicator for stenting in many institutions.[2,6] In our case, pressure gradients under local anesthesia were 23 and 17 mmHg before the first and second stenting, respectively. Under GA, the pressure gradients were 7 and 5 mmHg just before the first and second stenting procedures, respectively. Guo et al. reported manometry results of 32 IIH cases under awake settings and GA.[7] The mean venous pressures (MVPs) on the medial side of the stenotic site were non-significantly lower, and the MVPs on the distal side were significantly higher in the GA group. The pressure gradient was lower in the group under GA. However, in our patient, venous pressure was decreased on both the sides, and the pressure gradient was decreased under GA. Our patient's partial pressure of carbon dioxide (PCO_2) was 29.8 mmHg on arterial blood examination during stenting under GA; lowering of the MVP may be caused by hyperventilation accompanied by lowering ICP.[8] They recommend using venous pressure, measured under awake setting, instead of GA for stent indication of IIH.

SAS is a known complication after venous stenting for IIH. According to a review by Saber et al. on 24 articles, SAS occurred in 14% of the 473 patients after VSS.[5] However, there was no incidence of SAS in four of the 24 articles. However, one of the remaining 20 articles reported

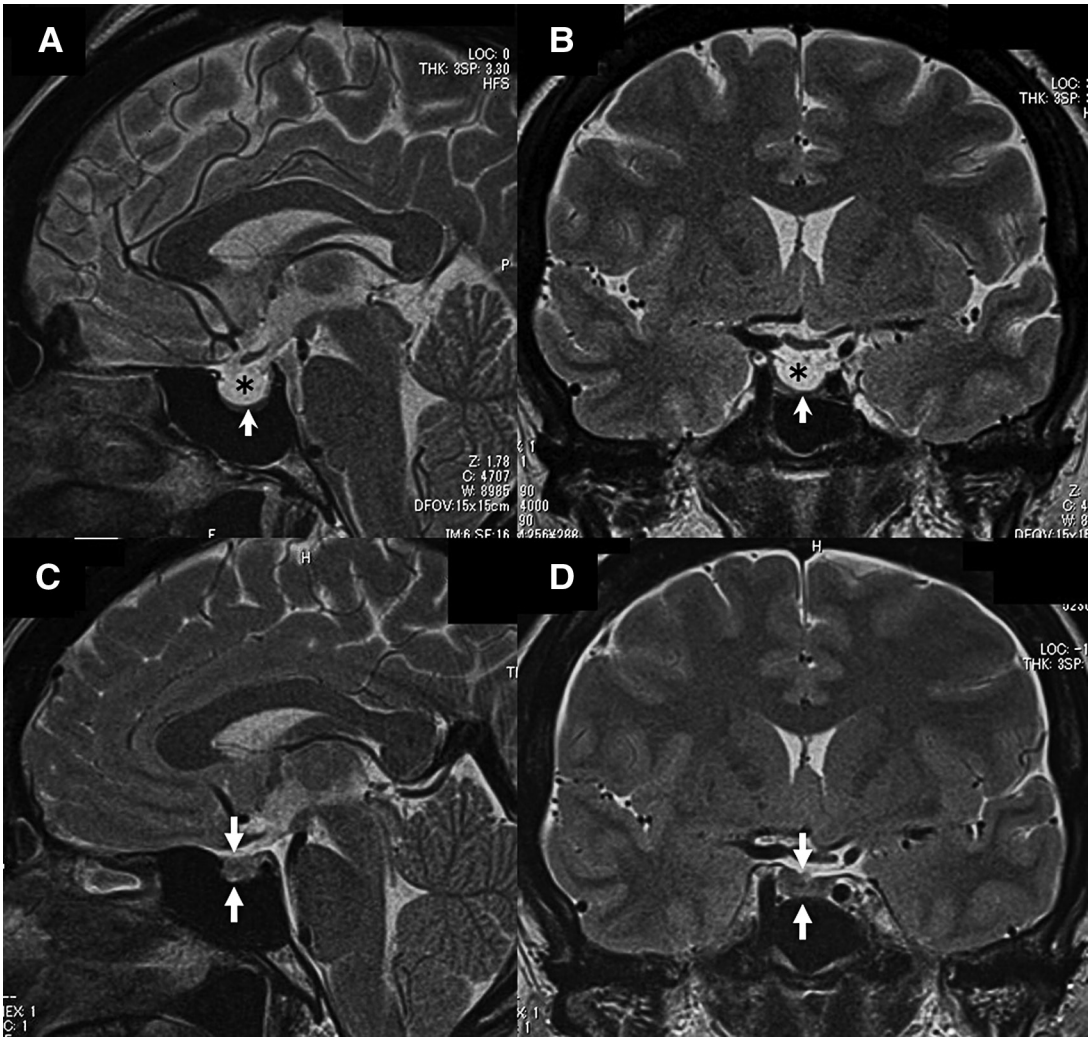

Fig. 3 (**A** and **B**) Sagittal and coronal views of MRI T2-WI before initial treatment reveal empty sella (asterisks) and a flattened pituitary gland (arrows). (**C** and **D**) Sagittal and coronal views of MRI T2-WI 10 months after second stent placement reveal increased height of the pituitary gland (arrows) and disappearance of the empty sella.

SAS in 36% of the patients.[5] Details on these variations are unknown; however, it is assumed that SAS developed owing to the use of stents that had an inappropriately large diameter, from the flow dynamic viewpoint.[9,10] As per empirical evidence, using longer stents at the initial stent placement is considered preferable to prevent SAS.[11–13]

Sheriff et al. reviewed the effects of the DAPT duration on stent survival, including SAS.[14] Although SAS occurred in 9% of the 325 cases, no significant association was noted between the DAPT duration and SAS incidence. To prevent stent-related thrombotic complications, DAPT is administered extrapolating from the evidence of carotid artery stenting.[14] Fargen et al. recommended administration of antiplatelet agents prior to stenting and for a minimum period of 3–6 months after stenting.[15] However, it remains unclear whether DAPT or single antiplatelet therapy is preferable over aspirin monotherapy, which reportedly caused thromboembolic complications in two patients in the past.[15,16] Saber et al. conducted a literature review on the regimen and duration of antiplatelet therapy.[5] In 21 articles, warfarin, aspirin, and clopidogrel were used after VSS. The majority of DAPT regimens included aspirin and clopidogrel for 3–6 months, followed by aspirin (100–325 mg/day) monotherapy for 6 weeks to 1 year or throughout life.[5]

Raper et al. reported that the prevalence rate for SAS after successful initial stenting is different based on the pattern of venous pressure gradient resolution.[17] They divided the pattern into the following three types. Type I is that higher MVP of upstream before stenting drops to the level of downstream. This type was observed in 18 of the 47 patients; SAS did not occur in these cases. Type II is the pattern wherein lower downstream pressure has risen up to

the level of the upstream pressure. This type was observed in seven patients; SAS occurred in two of the seven (28.6%) patients. Type III is the pattern wherein higher pressure decreased, lower pressure increased, and the pressure gradient was resolved. This type was observed in 22 patients, and SAS occurred in five of the 22 (22.7%) patients. Our patient belonged to the type III category under GA and was classified into no specific category under conscious sedation.

The mechanism of SAS as well as the etiology of IIH is not well elucidated. High ICP, sinus stenosis, elevated intravenous pressure, and water channel dysfunction affecting CSF circulation are believed to be involved in the pathophysiology of IIH. These factors are closely related to one another and create a vicious circle or maintain an equilibrium.[5] Studies have revealed two main mechanisms of IIH development, one of which is the extrinsic mechanism. This type of sinus stenosis is believed to be caused by compression due to raised ICP. From an anatomical perspective, the TS is vulnerable to collapse toward the lateral side due to lack of supportive structures, such as the septum and trabecula.[18] In patients with IIH, sinus stenosis occurs in the lateral side of the TS; thus, SAS may occur in the remaining parts of TS after stenting for the same reason.

In the present case, angiography revealed time-dependent change of the TS during SAS formation. The shape of SAS was not simply formed by the collapse of a floppy sinus immediately after initial stenting; rather, the narrowest portion of the stenosis seemed to develop near the edge of the stent over several months. The speed of SAS formation may vary. In cases of recurrent IIH (such as the present case) with no persistent signs and symptoms, SAS may form gradually. Stenting was believed to function by inhibiting the positive feedback loop for extrinsic stenosis, especially that caused by a mild increase in ICP.[19] Some improvement in ICP after VSS may lead to a mild increase in ICP, resulting in the gradual progression of restenosis in the remaining parts of TS. Therefore, additional stenting can be effective in cases of recurrent IIH by ensuring that the factors contributing to IIH are in equilibrium even before the completion of SAS formation.

Because normalization of ICP can reverse extrinsic type stenosis, CSF diversion is believed to be effective. In a study by Ducruet et al., eight out of 30 patients showed persistence or recurrence of symptoms after VSS, and five patients received CSF diversion at 1 week to 3 months after VSS.[20] Rohr et al. reported a case of IIH recurrence 1 week after VSS that formed SAS; they subsequently placed a ventriculoperitoneal shunt as increased ICP was the primary problem leading to venous sinus compression.[21] Our patient had an extrinsic stenosis, which was a long segment stenosis with obtuse margins and without any focal filling defect in the venous sinus lumen on MRV and angiography.[22] We did not opt for CSF diversion for TS stenosis and SAS because the patient developed severe headache and neck pain after collecting just 2–3 mL of CSF via lumbar puncture for examination.

Kumpe et al. reported the cases of patients with IIH accompanied by stenosis in the transverse–sigmoid junction who were treated with stenting; 10 of these 32 patients (31.3%) presented with SAS from the SSS to the TS.[13] They distinguished the remaining pressure gradient and recurrence after VSS by diagnosing hemodynamic failure and terming the disappearance of the pressure gradient as hemodynamic success. All the patients who experienced hemodynamic failure were women and had extrinsic stenosis, which is external compression of venous sinus determined on angiography. These patients were treated with restenting up to the SSS, as needed; all eight patients showed improvement in terms of papilledema with resolution of pressure gradient. Furthermore, according to the outcomes presented by Kumpe et al., the risk of SAS occurrence immediately after VSS should be considered owing to the fact that our patient presented with extrinsic stenosis.

In contrast, Ducruet et al. reported 60%–90% stenosis of the proximal (medial to the stent) SAS in five out of 30 patients.[20] Except for the case of one patient for whom ventriculoperitoneal shunt was performed, four of five cases were observed without additional treatment and showed symptom reduction during the follow-up period of 5.5–31 months under restenosis.[19] Thus, if proximal SAS is observed after initial stenting, the subsequent treatment strategy should be decided considering some conditions, such as signs, symptoms, and pressure gradient.

Conclusion

We reported on a patient with IIH that recurred and caused proximal SAS after initial stenting for TS stenosis and who required additional stenting. Retrospectively, angiography revealed progressive deformation of the remaining parts of TS after VSS. Predicting SAS based on the post-VSS clinical course and angiographic findings is important for selecting subsequent intervention and determining the appropriate timing of intervention. Clinicians should be aware that VSS is one of the treatment options for

restenosis (that may or may not be SAS), which can occur at any time in the remaining parts of TS. In such cases, a second stent placement may also be one of the options for administering effective and less invasive treatment.

Disclosure Statement

The authors declare no conflicts of interest.

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