CASE REPORT

Development of an intracranial dural arteriovenous fistula after venous sinus stenting for idiopathic intracranial hypertension

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SUMMARY

We report a case in which an intracranial dural arteriovenous fistula (DAVF) developed after endovascular treatment of a patient with idiopathic intracranial hypertension with venous sinus stenting (VSS). The pathogenesis may involve hemodynamic alterations secondary to increased poststenting venous sinus pressure, which may cause new arterial ingrowth into the fistulous sinus wall without capillary interposition. Despite administration of dual antiplatelet therapy, there may also be subclinical cortical vein thrombosis that contributed to DAVF formation. In addition to the aforementioned mechanisms, increased inflammation induced by VSS may upregulate vascular endothelial growth factor and platelet-derived growth factor expression and also promote DAVF pathogenesis. Since VSS has been used to obliterate DAVFs, DAVF formation after VSS may seem counterintuitive. Previous stents have generally been closed cell, stainless steel designs used to maximize radial compression of the fistulous sinus wall. In contrast, our patient's stent was an open cell, self-expandable nitinol design (Protégé Everflex). Neurointerventionalists should be aware of this potential, although rare complication of DAVF formation after VSS.

BACKGROUND

Stenting of the dural venous sinuses is becoming a popular treatment option for patients with medically refractory idiopathic intracranial hypertension (IIH).¹ Patients generally tolerate venous sinus stenting (VSS) without significant morbidity and mortality. However, data regarding long-term outcomes and complications associated with VSS is lacking. We report, to the best of our knowledge, the first case of dural arteriovenous fistula (DAVF) formation after VSS for a patient with IIH.

CASE PRESENTATION

A 22-year-old female college student (body mass index 32.24 kg/m²) presented with headaches, pulsatile tinnitus, peripheral vision loss, and papilledema. The patient's symptoms started 2 months prior to presenting to our cerebrovascular neurosurgery clinic. The patient could not identify inciting factors or notable events such as trauma that could have caused her symptoms. She did not regularly take any medication and denied significant medical or surgical history. Her family history was unremarkable. Aside

from papilledema on funduscopy, her neurological examination was non-focal.

INVESTIGATIONS

Lumbar puncture revealed an opening pressure of 36 cm H₂O, and she was diagnosed with IIH. The patient's symptoms persisted despite medical therapy with acetazolamide. Brain MRI and MR venography revealed a hypoplastic left transverse sinus (TS) and focal stenosis of the dominant right TS (figure 1A). Diagnostic cerebral angiography redemonstrated a focal venous stenosis at the right TS–sigmoid sinus (SS) junction from an arachnoid granulation, which was associated with a 10 mm Hg trans-stenotic pressure gradient (figure 1B).

TREATMENT

The patient underwent technically successful VSS of the superior sagittal sinus (SSS), right TS, and right SS with a construct of three overlapping Protégé Everflex (Covidien, Dublin, Ireland) stents. During the stenting procedure, a 5 mm Hg pressure gradient was detected across a narrowed segment of the posterior SSS, so the S1 segment of the SSS was included in the stent construct.² The post-stenting pressure gradient across the stent construct measured 4 mm Hg (figure 1C).

OUTCOME AND FOLLOW-UP

After 4 months of follow-up, the patient reported worsening headaches, pulsatile tinnitus, and peripheral vision. Ophthalmological exam revealed worsening papilledema. CT venography revealed worsening SSS stenosis adjacent to the initial stent construct. Cerebral angiography redemonstrated the stent-adjacent stenosis (SAS) in the posterior SSS, with a trans-stenosis gradient of 11 mm Hg. We previously reported a 14.9% rate of SAS (7/47) in a cohort of patients with IIH treated with VSS.³ In this case, the patient's SAS was treated with repeat VSS that extended the stent construct into the S2 segment of the SSS (figure 1D).³ During retreatment, we noted a de novo Borden type 1 DAVF which was not present on the previous angiogram. The DAVF had arterial supply from branches of the right meningohypophyseal and inferolateral trunks and venous drainage into the stented right TS/SS (figure 1E). Due to its antero-grade drainage pattern, the DAVF was classified as low risk for hemorrhage. Therefore, the lesion was initially managed conservatively.

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Figure 1 A 22-year-old female (body mass index 32.24 kg/m²) presented with worsening headaches, pulsatile tinnitus, peripheral vision loss, and papilledema. (A) Axial time-of-flight brain MRI shows hypoplastic left transverse sinus and focal stenosis of the dominant right transverse-sigmoid sinus junction from an arachnoid granulation (white arrow). (B) Venous manometry revealed an 11 mm Hg pressure gradient from the posterior superior sagittal sinus to the right sigmoid sinus. (C) Poststent DSA selective superior sagittal sinus injection shows three overlapping Protégé Everflex stents (8×40 mm, 8×40 mm, 10×40 mm) with arrows indicating the proximal and distal tines of the stent construct. There is a 4 mm Hg trans-stent pressure gradient from the superior sagittal sinus to the right sigmoid sinus. (D) Follow-up DSA shows widely patent stents, but stent-adjacent stenosis involving the posterior superior sagittal sinus (white arrow). We deployed a fourth overlapping stent (Cook Zilver 8×80 mm) across the stent-adjacent stenosis in the posterior superior sagittal sinus. The residual trans-stent pressure gradient was reduced to 1 mm Hg from the superior sagittal sinus to the right sigmoid sinus. (E) Right internal carotid artery DSA pre- and poststent placement shows a de novo Borden type 1 dural arteriovenous fistula fed by branches of the right meningohypophyseal and inferolateral trunks (white arrows) draining into the stented right transverse/sigmoid sinus.

DISCUSSION

DAVFs develop from pathological de novo arterial ingrowth into venous channels that bypass the interposed capillary network. The DAVF is not a direct AV shunt into the sinus lumen, but rather a connection between the dural arteries and dural veins within the venous sinus wall.^{4,5} Treatment of DAVFs includes endovascular embolization, surgical ligation, and stereotactic radiosurgery.^{6–8} Recently, authors have reported successful obliteration of DAVFs with VSS, in which the stent's radial compressive force occludes fistulous connections within the venous sinus wall.^{9–11} The inner endoluminal compression force on the sinus wall is related to the stent's radial force—which is dependent on the stent's mechanical characteristics. Closed cell, stainless steel stents such as the Boston Scientific Wallstent provide stronger homogenous radial force than their open cell, nitinol counterparts such as the stents used in this report.¹¹

We present the first case of DAVF formation after VSS for IIH; however, Lv *et al* described a similar case in which a patient with a direct carotid artery–cavernous sinus fistula (CCF) developed an indirect CCF (ie, a DAVF) after treatment with a covered stent in the cavernous segment of the internal carotid artery.¹² The authors hypothesized that stent placement caused hemodynamic alterations that promoted CCF revascularization via small branches of the meningeal supply.¹² We also hypothesize that our patient's DAVF formation may be explained by poststent hemodynamic alternations, which may in part be related to venous pressure elevations that diminished cortical drainage into the stented segment of the dural sinus. The prestenting SS mean venous pressures (MVP) were 3 and 9 mm Hg with conscious sedation and general anesthesia, respectively. After stent placement, the pressure gradient decreased, but poststenting SS MVP increased to 12 mm Hg under general anesthesia. The elevated SS MVP may have reduced cortical venous inflow, which then promoted pathogenesis of the DAVF. The patient's follow-up imaging showed that the stent was not undersized to the parent sinus wall nor was ovalization observed (figure 1D). In spite of this, the mechanical characteristics of the Protégé Everflex stent (open cell, nitinol) did not provide a sufficient high radial compressive force to occlude new fistulous connections in the dural sinus wall.

If appropriate antiplatelet therapy is not administered during the time required for epithelialization of the stent by endothelial cells, cortical vein occlusion may occur.¹¹ Although our patient was maintained on dual antiplatelet therapy with therapeutic Aspirin and P2Y assay levels, subclinical cortical vein thrombosis may have contributed to DAVF pathogenesis. It is possible that the mesh of the nitinol stent bridged the ostia of cortical veins draining into the stented TS/SS. This foreign body may have induced cortical vein occlusion and subsequent DAVF formation. Although cortical vein occlusion was not observed on follow-up angiography, this cannot be excluded since transient thrombosis and recanalization may have occurred.

Cardiac literature describes neointimal angiogenesis, inflammation, and increased vascular endothelial growth factor (VEGF) and platelet-derived growth factor (PDGF) within human coronary arteries after stenting.¹³ The degree of VEGF and PDGF expression after VSS is uncertain, but likely increased especially in cases of poststent neointimal hyperplasia. We hypothesize that increased inflammation induced by VSS may upregulate VEGF and PDGF expression, and this mechanism may have promoted DAVF pathogenesis in the patient.

We describe a novel case of the development of a de novo DAVF following VSS for the treatment of IIH. The pathogenesis of our patient's DAVF may involve poststenting hemodynamic

alterations due to refractory elevated poststenting venous pressure, subclinical cortical vein thrombosis, and poststent inflammation with upregulated VEGF and PDGF expression. Neurointerventionalists should be aware of the possibility of this very rare complication of DAVF formation after VSS.

Contributors All authors made substantial contributions to the conception, data acquisition and analysis, interpretation or drafting of this research article. We approve this final version to be published.

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