Abstract

We report a case of a 54-year-old man with an occluded straight sinus and dural AV-fistula. The patient was treated with PTA dilatation and stenting of the occluded straight sinus. A review of literature revealed no similar therapy has to date been described.

Introduction

Cerebral dural venous sinus thrombosis (CVST) is the cause of less than 1% of all strokes and results in an overall mortality rate of 4% [1-2]. A common, effective and safe treatment in the management of CVST is systemic anticoagulation. In a study by Coutinho et al., a 13% reduction in death and dependency was found among heparin-treated patients without symptomatic intracerebral haemorrhage (ICH) [3]. However, 8% of all heparin-treated patients die and 13% score ≥2 on the modified Rankin Scale. Several prognostic factors play a role in poor outcome: patient age, sex (male), coma, mental status disorder, thrombosis of the deep cerebral venous system, ICH, malignancy and central nervous system (CNS) infection.

Endovascular thrombolysis treatment should be considered especially with high-risk patients. The first report of mechanical thrombectomy was published by Opatowsky et al. in 1999 [4]. More recently, different thrombectomy devices have offered new choices for an endovascular thrombolysis approach when treating high-risk patients. Several case reports on endovascular thrombolysis using mechanical or pharmaceutical thrombolytic devices have been published. In some cases, stents are used to open occluded dural sinuses [5] such as in cases of idiopathic intracranial hypertension (IIH).

Technique

The patient in this case study was a 54-year-old forest worker who had previously been diagnosed with high blood pressure but was otherwise healthy and active. Over a period of a few months, he had suffered from failing memory, lethargy and somnolence. During the last two weeks prior to treatment, his symptoms dramatically worsened and during the last couple of days, he mainly sat still at home. He was found silent and rigid.

The patient was taken to the local hospital. At the hospital, he could only give one-word answers and he suffered from right hemiparesis. Later, he had convulsions. Emergency computerised tomography (CT) showed a small ICH near the head of the left caudate nucleus and a subarachnoid haemorrhage (SAH) in the basal cisterns (Fig. 1a). The thalami were hypodense (Fig 1b). Pre-epanutin and acyclovir medication was started in the referring hospital. The patient was then intubated and transferred to our institute for additional examinations.
MRI imaging showed that both thalami and the left basal ganglia were oedematous with petechial haemorrhage (Fig. 1 b-c). Diffusion imaging showed no infarction. The straight sinus was not shown and the vein of Galen was prominent. The left transverse sinus was hypoplastic, presumably as a normal variant. Otherwise, the venous sinuses were open.

In digital subtraction angiography (DSA), a dural AV-fistula (DAVF) into the vein of Galen from a minor posterior meningeal branch of the left vertebral artery was found. The straight sinus was occluded and the prominent vein of Galen was drained pathologically anteriorly (Fig. 2a).

We changed to the venous approach to catheterise the right transverse sinus and proceeded into the vein of Galen through the closed straight sinus. First, we tried repeatedly to aspirate the thrombus with a distal access catheter (DAC), (Concentric Medical, Mountainview, California, USA), and a Trevo mechanical thrombectomy device (Concentric Medical, Mountainview, California, USA) without success. Percutaneous transluminal angioplasty (PTA) of the straight sinus showed a stenosis between the vein of Galen and the straight sinus and another stenosis in the middle of the straight sinus (Fig. 2b). The PTA balloon was pulled back when dilated without capturing any thrombus material. It appeared that there was only a chronic organised thrombus and a stenosis of the straight sinus. After PTA, moderate flow was achieved in the straight sinus, but the DAVF remained open.

Two days after PTA, we tried to close the DAVF. The straight sinus remained open, and the retrograde venous flow from the vein of Galen was decreased. The basis of the feeder artery was very torturous and when trying to catheterise this under a 1.5 mm artery with a 0.012" Sonic (BALT, Montmerci, France) catheter, we dissected it right after its basis. A post-treatment DSA series showed that this feeder artery was occluded.

In MRI imaging 9 days after the final DSA, the oedema seen in the thalamic and basal ganglia was diminished, but the feeder of the DAVF was reopened. The straight sinus remained open and no definitive infarcts were seen.

Small molecular heparin medication was started at 60 mg x 2. The patient’s condition slowly improved over a period of some weeks. The mini mental state examination (MMSE) was 25/30, but 2.5 months after the initial endovascular treatments, the patient’s condition began to deteriorate. Right hemiparesis progressed. The patient’s cognition and general condition weakened and the MMSE dropped to 12/30. In repeated MRI imaging, the straight sinus appeared re-occluded. The oedema was increased and continued from the thalami to the left crus cerebri and periaqueductal parenchyma and to the posterior limb of the left capsula interna. Diffusion imaging showed no irreversible ischemic areas.

We recanalised the occluded straight sinus again and dilated it with a PTA balloon (Fig 2c). Two Enterprise (Codman, Raynham, Massachusetts, USA) stents were delivered telescope-like from the vein of Galen to the posterior part of the straight sinus. Post-stenting DSA showed good flow in the straight sinus (Fig. 2d). The DAVF was still open. While trying to catheterise the feeder of the DAVF, the microcatheter perforated the tiny feeder. Because clopidogrel (75 mg x 1), aspirin (100 mg x 1) and heparin infusion (APTT 60-100) had been started to prevent stent thrombosis, it was crucial to embolise the leaking small artery. This was achieved with one 1.5 x 20 mm platinum coil. A post-procedural series showed normal flow in the vertebral artery and the straight sinus. The pathological retrograde venous flow seen before was now minimal. The DAVF was closed.
Five days after the last endovascular treatment, CT imaging still showed the same thalamic hypodensity seen in previous CT scans. The straight sinus was open. Nine months after the first and 6.5 months after the second endovascular sessions, CT imaging of the head (Fig. 3) showed normal symmetric thalami and basal ganglia and the straight sinus remained open. The patient felt very well and, for example, exercised by taking daily 10 km walks. The patient planned to return to work as a logger and has had his motorcycle driving license reinstated.

Discussion
In cases of dural sinus thrombosis, systemic heparinisation is the front-line treatment. If, however, a patient has poor prognosis, more invasive techniques such as mechanical thrombectomy should be considered. Here, we present to the best of our knowledge, the first case of stenting of the straight sinus due to its thrombus. The clinical recovery was successful in long-term follow-up. As venous hypertension is seen a major causative factor in the formation of a non-traumatic dural AVF [6-7], it might seem rational to focus on the elimination of this factor. We were not able to successfully perform mechanical thrombectomy by aspiration or by using a thrombus-retrieving device in this medical emergency case. Instead, local PTA was performed resulting in some opening of the straight sinus lumen. We did not try local thrombolysis because the thrombus was very solid and resistant to its mechanical fragmentation or vessel dilation. Furthermore, the patency of the sinus lumen was achieved only after stent implementation.

The concomitant - or subsequent - arterial feeder was still open 2 days after the primary straight sinus PTA session. In a second session, the arterial feeder of the AV-fistula was occluded, not tentatively, but due to its dissection. After 2.5 months, however, the feeder appeared to be reopened again and, as the patient was on heparin with a recent history of the arterial dissection, the feeder's closure was regarded as mandatory and was finalised with a coil. In the second therapeutic session 2.5 months after the primary session, a telescope-like stenting of the sinus after a repeated dilation was performed. As a result, both the venous and the arterial pathology disappeared followed by a favourable clinical recovery. The draining of an AV-fistula directly to the deep venous system without cortical reflux may be regarded as benign considering there was only risk of haemorrhage. The increased intracranial pressure due to compromised venous outflow, however, resulted in ischaemia and quite rapid clinical deterioration, though permanent ischaemia did not develop.

After the endovascular sessions, the patient received further treatment at the referring hospital. Our recommendation was that Marevan therapy with its laboratory monitoring should continue for 6 months with Plavix 75 mg daily and ASA 100 mg daily. As the feeding artery of the AV-fistula was closed at its proximal segment, recanalisation remains a risk. However, if the straight sinus remains open, it will not stay as a predisposing factor for fistula formation. It is also probable that rethrombosis of the straight sinus would be followed by a rapid and drastic clinical exacerbation as seen in the primary phase. So far, we have not performed any catheter angiographies as the straight sinus patency could be properly visualised with cross-sectional imaging. This remains the case as long as the patient`s clinical condition remains uncomplicated, which has been the case for the past 3.5 years.

Conflict of interest
We declare that we have no conflict of interest.
References


Figures

**Figure 1** - In the preliminary CT image (a) there is blood in the quadrigeminal/ambient cisterns and in the third ventricle, a focal haemorrhage is seen in the caput of the left caudate nucleus. The thalami, especially in the left, are hypoattenuated (b) due to oedema. There is a signal loss seen in the SWI image (c) due to the focal haemorrhage in the SWI, and also scattered signal loss areas due to the deoxygenated blood in the veins and probably due to petechial haemorrhages. The oedematous areas of the central gray matter are well delineated in the MR/Flair image (d).
Figure 2 - DSA shows DAVF into vein of Galen and pathologic venous drainage anteriorly (a). PTA balloon reveals short stricture of the straight sinus (b). Contrast media injected into the straight sinus after recanalisation and PTA (c). Pathologic anterior venous drainage is dramatically reduced and ordinary flow in the straight sinus is achieved after stenting (d).
Figure 3 - In the follow-up CT image (a) the oedematous hypodensity has well resolved as is the focal haemorrhage - note the stent markers. The sagittally reconstructed CTA image (b) shows the open straight sinus and the stent markers in the follow-up.