CASE REPORT

Delayed postoperative dural arteriovenous fistula presenting with progressive dementia

Tetsuhiro Higashida

Department of Neurosurgery,
Yokohama City University,
School of Medicine,
Yokohama, Kanagawa, Japan

Correspondence to
Dr Tetsuhiro Higashida,
tets018@gmail.com

Accepted 19 November 2015

SUMMARY

A 64-year-old woman underwent right suboccipital craniotomy to treat spontaneous cerebrospinal fluid (CSF) otorrhoea. Although the CSF leak was cured, the patient developed pulsatile tinnitus, ambulatory disturbance and progressive dementia. Four years after the surgery, she presented with generalised seizure. Cerebral angiography showed a dural arteriovenous fistula at the right transverse-sigmoid sinus, which was associated with sinus occlusion, retrograde blood flow in the sinus and diffuse intracranial cortical venous reflux. A combination of endovascular transarterial embolisation and transvenous embolisation with direct sinus puncture was performed. After the procedure, the patient's mental status and cognitive function improved significantly.

BACKGROUND

Although the aetiology and pathogenesis of dural arteriovenous fistulas (AVFs) remain to be fully elucidated, acquired dural AVFs have been reported to develop following craniotomy.^{1–4} The present study is a rare case of postoperative dural AVF occurring at the site of craniotomy for the treatment of cerebrospinal fluid (CSF) leak, and some operative manoeuvres that might facilitate the development of dural AVF are discussed.

CASE PRESENTATION

A 64-year-old woman underwent surgical treatment of spontaneous right CSF otorrhoea at another institute. CSF appeared to leak into the right mastoid air cells through a partial bony defect of the right pyramidal bone (figure 1A). A dural hole, which allowed leakage of the CSF, was located at the pre-sigmoid dura just behind this bony defect (figure 1B). After extended suboccipital craniotomy was

performed and the CSF leakage point identified, the dural hole was sutured primarily. Then a large vascularised muscular flap, which was mainly fed by the occipital artery, was used to fill in the epidural space between the bony defect and the closed dural hole to provide a further seal. The exposed sigmoid sinus was also covered by this vascularised flap. After the surgery, the CSF leak was cured; however, the patient gradually developed pulsatile tinnitus, ambulatory disturbance and progressive dementia, which finally resulted in the need for persistent nursing care.

Four years after treatment of the CSF leak, she was transferred to our hospital, presenting with generalised seizure. She turned out to have a massive dural AVF at the right transverse-sigmoid sinus (figure 2A–D). The arterial blood supply came from multiple branches of the right external carotid artery, which connected directly to a shunting point located at the lateral right transverse sinus. The right sigmoid sinus was totally occluded and retrograde venous flow associated with diffuse intracranial cortical venous reflux was noted. Treatment of the dural AVF was indicated because her symptoms, including cognitive dysfunction and generalised seizure, were likely to be caused by chronic intracranial venous hypertension due to the dural AVF. A combination of endovascular transarterial embolisation and transvenous embolisation with direct sinus puncture was performed. After this combined embolisation procedure, the AVF was completely obliterated.

OUTCOME AND FOLLOW-UP

At 6-month follow-up, the patient's neurological status improved significantly. She was still mildly disoriented, but became almost independent. Follow-up angiography showed stable occlusion of the AVF (figure 3A, B).

DISCUSSION

Dural AVFs have various clinical manifestations, depending on the anatomical location and the venous drainage pattern, but rarely present as dementia.^{5–6} The presumed mechanism of progressive dementia from dural AVF appears to be venous congestive encephalopathy caused by intracranial venous hypertension. As evolution of radiological equipment and embolisation devices has occurred, selective endovascular embolisation has become the primary treatment method for dural AVF. This procedure can achieve immediate obliteration of arteriovenous shunt as well as venous hypertension,

Figure 1 Three-dimensional CT showing the right pyramidal bony defect (arrow) (A). Axial T2-weighted MRI showing meningeal herniation through the right pyramidal bony defect and fluid collection in the right mastoid air cells (arrow) (B).

To cite: Higashida T. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-212874

Figure 2 Axial contrast-enhanced CT showing right suboccipital craniotomy (arrow) (A) and numerous dilated intracranial veins (B). Anteroposterior (C) and lateral (D) view of angiography showing the arteriovenous fistula between the branches of the right external carotid artery and the right transverse sinus. Note the right sigmoid sinus is not visualised due to occlusion.

which possibly results in the improvement of cognitive dysfunction.

Delayed postoperative dural AVFs have rarely been proven angiographically. Six patients with delayed development of dural AVF involving a venous sinus at the site of craniotomy have been reported, including the present case.^{1 2 4 7} All reported cases were AVFs involving the transverse-sigmoid sinus and five cases were AVFs that developed after suboccipital craniotomy.

Venous sinus thrombosis is considered to be one of the important causative factors in the development of dural AVF.^{1-3 5} Venous hypertension following sinus thrombosis can induce physiological dural arteriovenous channels to open and communicate with the patent portion of the sinus, ultimately generating a dural AVF. In surgery with suboccipital craniotomy, a transverse-sigmoid sinus is often exposed and manipulated. These procedures can lead to desiccation or injury of the dura surrounding the sinus followed by thrombus formation in the sinus.^{2 8}

Another proposed mechanism of postoperative dural AVF is the development of neovessels from extracranial tissues. In the healing process after craniotomy, angiogenesis can occur from the apposed hypervascular tissues, such as the scalp and muscles. These neovessels grow into the intracranial space and provide additional arterial supply to the dura, which can then develop a dural AVF.^{1 2 9} In surgery with suboccipital craniotomy, extra drilling of the bone is often performed to obtain an adequate surgical field. This wide bony defect can allow extracranial tissues to come into close contact with the dura, which could be the reason for the high incidence of postoperative dural AVF after suboccipital craniotomy.

In the present case, a thick vascularised muscular flap was packed into the epidural space to achieve a secure seal for the CSF leak. This flap broadly covered the sigmoid sinus, potentially compressing the sinus and inducing thrombosis. Additionally, the close contact of the vascularised tissues to the dural surface of the venous sinus might facilitate recruitment of new dural vasculature and lead to the progressive development of AVF.

Learning points

- ▶ Suboccipital craniotomy is the most common approach that can develop a dural arteriovenous fistula (AVF); therefore, surgeons must keep in mind that extra manipulation of the exposed venous sinus and apposition of the vascularised tissues on the sinus should be withheld to reduce the risk of postoperative dural AVF.
- ▶ Dural AVF should be considered as a differential diagnosis for progressive neurological deterioration in a patient with a history of craniotomy. Careful postoperative clinical follow-up and timely radiological assessment are crucial in early diagnosis.
- ▶ Symptomatic dural AVF associated with intracranial venous hypertension should be promptly treated by appropriate endovascular embolisation because the symptoms are potentially reversible.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

Figure 3 Anteroposterior (A) and lateral (B) view of angiography after the embolisation showing complete obliteration of the arteriovenous fistula.

REFERENCES

- 1 Nabors MW, Azzam CJ, Albanna FJ, *et al.* Delayed postoperative dural arteriovenous malformations. Report of two cases. *J Neurosurg* 1987;66:768–72.
- 2 Sasaki T, Hoya K, Kinone K, *et al.* Postsurgical development of dural arteriovenous malformations after transpetrosal and transtentorial operations: case report. *Neurosurgery* 1995;37:820–4; discussion 4–5.
- 3 Chen L, Mao Y, Zhou LF. Local chronic hypoperfusion secondary to sinus high pressure seems to be mainly responsible for the formation of intracranial dural arteriovenous fistula. *Neurosurgery* 2009;64:973–83; discussion 83.
- 4 Kim SH, Chang WS, Jung HH, *et al.* Delayed dural arteriovenous fistula after microvascular decompression for hemifacial spasm. *J Korean Neurosurg Soc* 2014;56:168–70.
- 5 Ma C, Lu Q, Shi W, *et al.* Diagnosis and treatment of a dural arteriovenous fistula presenting with progressive parkinsonism and dementia: a case report and literature review. *Exp Ther Med* 2015;9:523–6.

- 6 Waragai M, Takeuchi H, Fukushima T, *et al.* MRI and SPECT studies of dural arteriovenous fistulas presenting as pure progressive dementia with leukoencephalopathy: a cause of treatable dementia. *Eur J Neurol* 2006;13:754–9.
- 7 Tanabe J, Ishikawa T, Moroi J, *et al.* [Development of dural arteriovenous fistula following lateral suboccipital craniotomy]. *No Shinkei Geka* 2013;41:711–16.
- 8 Keiper GL Jr, Sherman JD, Tomsick TA, *et al.* Dural sinus thrombosis and pseudotumor cerebri: unexpected complications of suboccipital craniotomy and translabyrinthine craniectomy. *J Neurosurg* 1999;91:192–7.
- 9 Uranishi R, Nakase H, Sakaki T. Expression of angiogenic growth factors in dural arteriovenous fistula. *J Neurosurg* 1999;91:781–6.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow