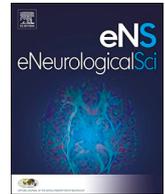




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Case report

Development of a dural arteriovenous fistula subsequent to cerebral venous thrombosis by venous hypertension

Min Kyoung Kang^a, Young-Dae Cho^b, Hyun-Seung Kang^c, Keun-Hwa Jung^{a,*}^a Department of Neurology, Seoul National University Hospital, 101 Daehak-ro, Jongno-gu, Seoul 03080, Republic of Korea^b Department of Radiology, Seoul National University Hospital, 101 Daehak-ro, Jongno-gu, Seoul 03080, Republic of Korea^c Department of Neurosurgery, Seoul National University Hospital, 101 Daehak-ro, Jongno-gu, Seoul 03080, Republic of Korea

ARTICLE INFO

Keywords:

Cerebral venous thrombosis
Dural arteriovenous fistula

ABSTRACT

Dural arteriovenous fistulas (dAVF) refer to an aberrant connection between an artery and a vein within the dura. Although the pathogenesis of dAVF is unclear, a link to cerebral venous thrombosis (CVT) has been posited though not fully identified. The current case is the first report demonstrating dAVF formation following CVT according to dynamic changes in the intracranial pressure and venous drainage pattern. This observation provides insight into the pathophysiological association between dAVF and CVT. A 60-year-old woman presented with acute onset of a severe headache and first-onset seizure. Initial magnetic resonance imaging showed an extensive thrombosis in the cerebral venous sinuses. However, there was no evidence of any vascular malformation. Eight months later, she reported dizziness, visual obscurations and pulsatile tinnitus. Follow-up digital subtraction angiography showed multiple dAVFs. Endovascular treatments for the dAVF embolization was performed twice, resulting in the obliteration of the dAVF along with the resolution of her optic disc edema, visual obscurations and tinnitus. The degree and pattern of the venous pressure, not hypoxia-induced growth factors, are the key pathogenic mechanisms of dAVF following CVT. Oral anticoagulants and good adherence do not ensure the prevention of dAVF. Thus, careful clinical observation and follow-up examinations are recommended.

1. Introduction

A dural arteriovenous fistula (dAVF) is a type of vascular malformation characterized by an aberrant connection between an artery and a vein within the dura. Although the pathogenesis of dAVF is not well known, a link to cerebral venous thrombosis (CVT) has not been identified with regard to any cause-and-effect relationship, and its direction, between CVT and dAVF.

Few studies have reported cases involving both dAVF and CVT. To the best of our knowledge, the current case is the first report that demonstrates the formation of dAVF following CVT according to dynamic changes in the venous drainage pattern. This observation sheds light on the association between the pathophysiology of dAVF and CVT and provides an effective follow-up strategy after CVT.

2. Case description

A 60-year-old woman presented with acute onset of a severe headache, visual obscuration and first-onset seizure. She had a history of

hypertension and dyslipidemia but no prior illnesses. A fundus examination showed bilateral papilledema. Initial magnetic resonance imaging showed extensive thrombosis (Fig. 1). We found no source of thrombophilia. She was treated with Warfarin and Levetiracetam. During her in-hospital period, she reported that her headaches had improved gradually but that her papilledema had become aggravated (Fig. 2). Digital subtraction angiography (DSA) was done. The venous drainage by the right anterior system showed a remarkable increase compared to earlier imaging. No findings suggestive of vascular malformation were detected. She was discharged, and her initial treatment was maintained.

Eight months later, she reported visual obscurations and pulsatile tinnitus. Follow-up DSA showed multiple dAVFs and progression of the superior sagittal sinus thrombosis (Fig. 3). Endovascular treatments of the embolization was performed twice, resulting in the obliteration of the dAVF along with the resolution of her optic disc edema, visual obscuration and tinnitus.

Written informed consent was obtained from the patient for publication

* Corresponding author.

E-mail address: jungkh@gmail.com (K.-H. Jung).<https://doi.org/10.1016/j.ensci.2018.11.015>

Received 1 November 2018; Accepted 17 November 2018

Available online 22 November 2018

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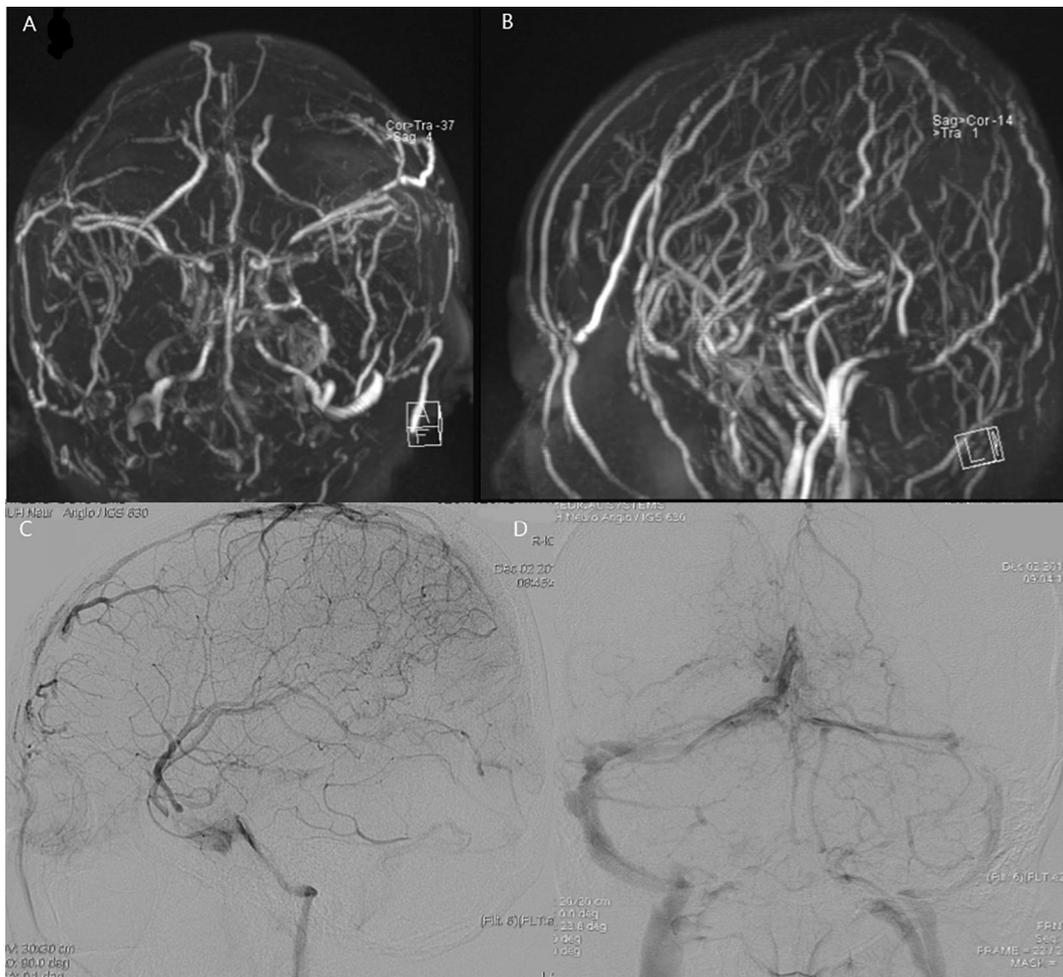


Fig. 1. Magnetic resonance venography at the time of presentation shows extensive thrombosis in the superior sagittal sinus, straight sinus, right transverse sinus, and right jugular vein(A, B). Right internal carotid angiogram showed an occlusion of the posterior one-fourth of the superior sagittal sinus and a drainage pattern into the anterior venous system(C). Vertebral angiogram showed a poor visualization of the left transverse sinus(D).

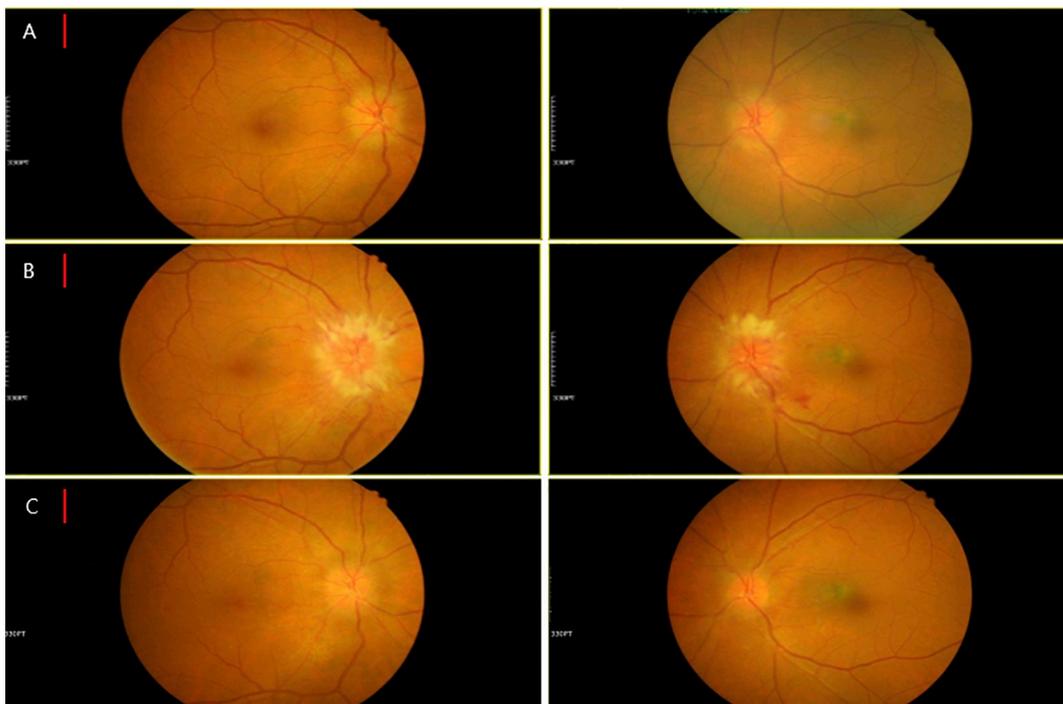


Fig. 2. In the fundoscopic exam at presentation(A), bilateral papilledema was noted. After three weeks(B), the bilateral papilledema had worsened despite the improvement of her symptoms. After the second AVF embolization, her papilledema was resolved with improvement of visual obscuration(C).

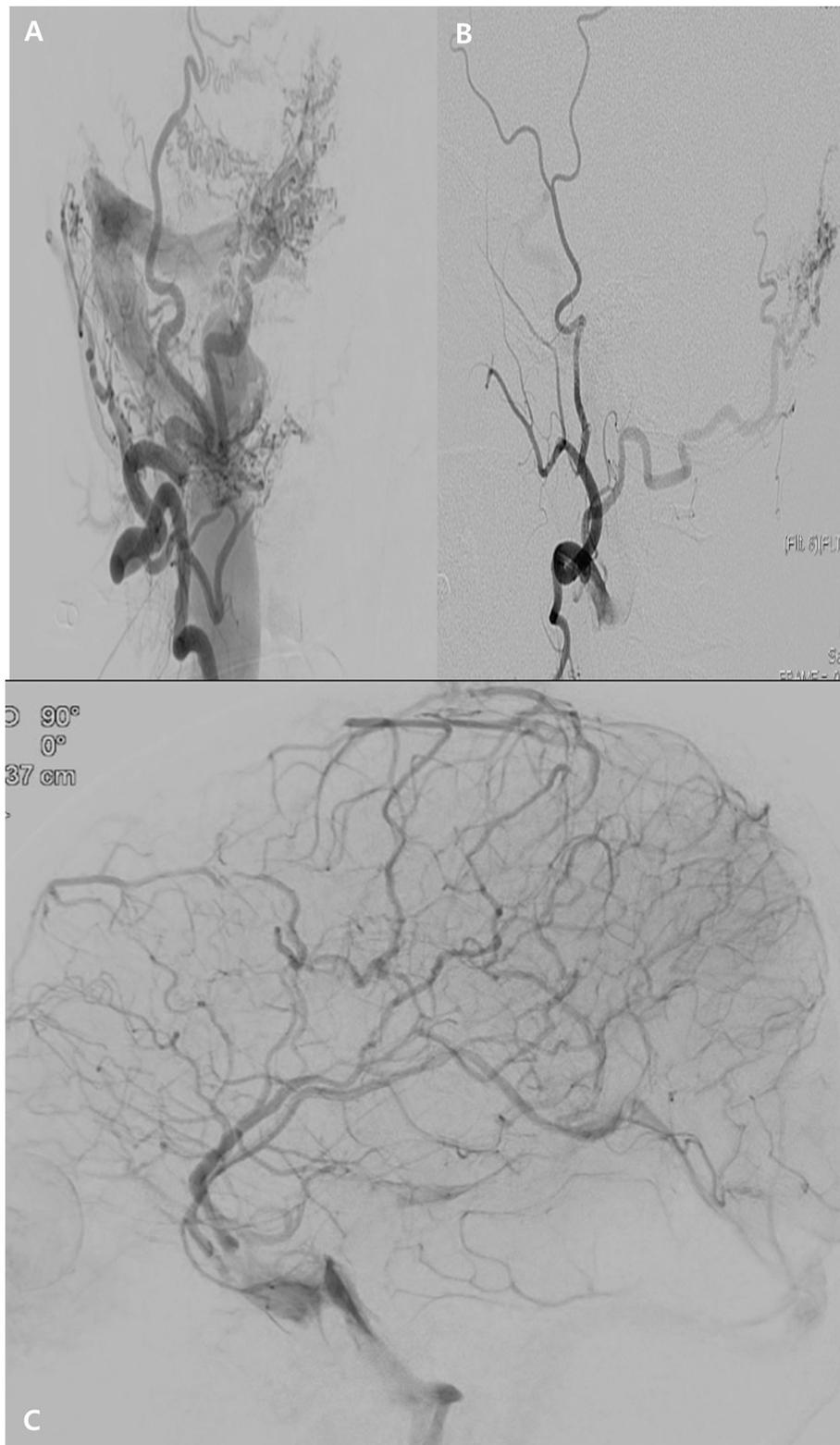


Fig. 3. Nine months after initial presentation, right external carotid angiogram showed multiple dAVFs in the torcular herophili, right transverse sinus and right sigmoid-jugular bulb(A). Left external carotid angiogram showed torcular dAVF(B). There was a larger amount of veins drained into the anterior venous system compared to Fig. 1-C(C).

3. Discussion

The common feature of dAVF following CVT is its development adjacent to a thrombosed sinus [1,2]. Hypoxia-induced angiogenesis or a reopening of preexisting channels by venous hypertension have been suggested as pathogeneses of dAVF following CVT [3,4]. However, in our case, thrombosis in the straight sinus, right transverse sinus, and right jugular vein, where the dAVF developed thereafter had resolved. Meanwhile, there was a drainage shift to the right anterior venous system, which possibly caused persistent hypertension in the right cortical veins. The local venous hypertension may have contributed to the development of dAVFs, unilateral pulsatile tinnitus and persistence of the papilledema. Improvement of general venous hypertension through the right anterior venous system may have relieved headache. This case could not be fully explained by the hypoxia-induced angiogenesis/reopening model, suggesting that the venous pressure pattern and dynamics are the key pathogenic mechanisms of dAVF following CVT.

Treating CVT with an oral anticoagulant has been recommended. However, this case shows that oral anticoagulants and good adherence alone do not ensure the prevention of dAVF following CVT. Thus, a clinical evaluation to assess history of unilateral pulsatile tinnitus, a venous flow status evaluation by angiogram and a regular follow-up fundoscopic exam are recommended for CVT patients. This

strategy would make effective and timely intervention possible when dAVF develops during the treatment of CVT with an oral anticoagulant.

Conflicts of interest

The authors declare that they have no financial or other conflicts of interest in relation to this research.

Declarations of interest

None

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