

Development of a sigmoid sinus dural arteriovenous fistula secondary to sigmoid sinus thrombosis after resection of a foramen magnum meningioma: illustrative case

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BACKGROUND The precise etiology of dural arteriovenous fistula (DAVF) is still unknown. The authors reported a case of delayed postoperative sigmoid sinus (SS) DAVF secondary to SS thrombosis after resection of a foramen magnum meningioma through a suboccipital craniotomy.

OBSERVATIONS The authors visualized the clear architecture of the DAVF using fusion three-dimensional computer graphics (3DCG) images reconstructed from multimodal imaging studies. These fusion 3DCG images revealed that the feeders of the DAVF had connected through neovascularization to the SS at the previous thrombus site. The authors also reviewed previously reported cases of DAVFs that developed after craniotomy.

LESSONS This study indicated that SS stenosis and occlusion with sinus thrombosis are possible risk factors for delayed postoperative DAVF that demand special consideration.

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KEYWORDS dural arteriovenous fistula; foramen magnum meningioma; sinus thrombosis; sigmoid sinus; fusion three-dimensional computer graphics images

Dural arteriovenous fistula (DAVF) is an uncommon vascular entity that consists of direct pathological connections between meningeal arteries and dural venous sinuses or leptomeningeal veins. DAVFs represent 10% to 15% of all intracranial vascular malformations.¹ The etiology of DAVFs is still uncertain, but they are reportedly associated with venous sinus thrombosis, trauma, and previous craniotomy, among other factors.²⁻⁴ Here, we present a case of sigmoid sinus (SS) DAVF that developed secondary to SS thrombosis after resection of a foramen magnum meningioma as well as a review of the relevant literature.

Illustrative Case

A 63-year-old woman presented with clumsiness and numbness of both upper limbs. Magnetic resonance imaging (MRI) showed a

foramen magnum meningioma (Fig. 1A). Preoperative cerebral angiography showed that the occipital artery (OA) and ascending pharyngeal artery (APA) were the arterial feeders (Fig. 1B and C). The presence of stenosis of the SS or intracranial vascular malformations was not confirmed (Fig. 1D). Considering that the meningioma was symptomatic, we performed a tumor resection via a right suboccipital craniotomy. Gross-total resection (resection extent: Simpson grade II) was achieved, and the histopathological diagnosis was meningothelial meningioma (World Health Organization grade 1). During craniotomy, bleeding from the SS was encountered, and hemostasis was achieved by compression. Although the patient recovered well without any new neurological deficits, contrast-enhanced MRI demonstrated occlusion (Fig. 2A) of the right SS and thrombosis (Fig. 2B) on postoperative day 1. Because this lesion was asymptomatic, the patient was

ABBREVIATIONS 3DCG = three-dimensional computer graphics; APA = ascending pharyngeal artery; CVR = cortical venous reflux; DAVF = dural arteriovenous fistula; MRA = magnetic resonance angiography; MRI = magnetic resonance imaging; OA = occipital artery; SS = sigmoid sinus.

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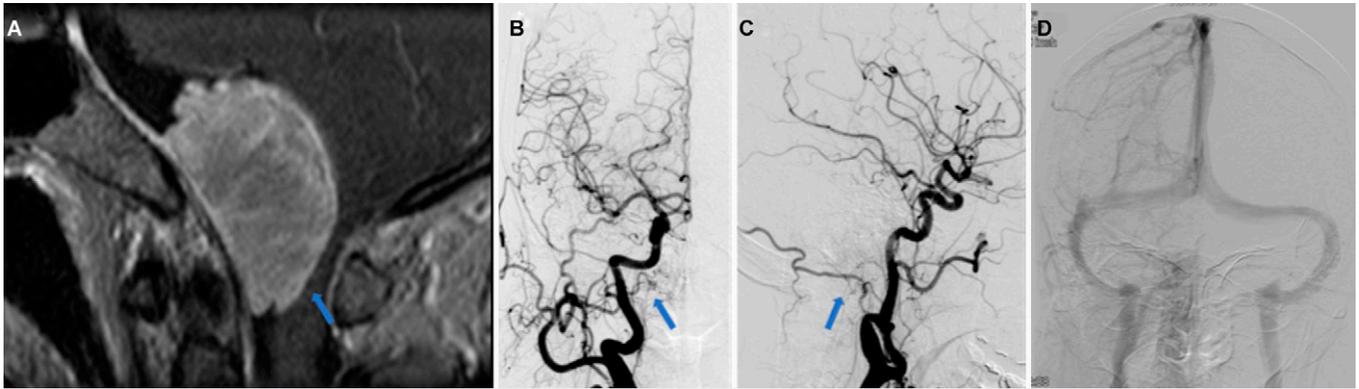


FIG. 1. Preoperative imaging. **A:** Sagittal gadolinium-enhanced MRI before tumor resection. An extraaxial tumor with enhancement on the foramen magnum was detected (*blue arrow*). **B:** Anteroposterior view of digital subtraction angiography of the right common carotid artery before surgery. The right APA (*blue arrow*) was feeding the tumor. **C:** Lateral view of digital subtraction angiography of the right common carotid artery before surgery. The right OA (*blue arrow*) was feeding the tumor. **D:** Anteroposterior view of digital subtraction angiography of the right common carotid artery before surgery. No obvious vascular malformations or stenosis of the sigmoid sinus was detected.

managed conservatively without the use of heparin. No intracranial vascular malformations had been detected at this point. At 47 months postoperatively, MR angiography (MRA) incidentally showed the right transverse sinus and SS, which were not visualized immediately after surgery, and increased abnormal blood vessel growth (Fig. 2C and D). Cerebral angiography at 49 months postoperatively revealed the development of an SS DAVF with occlusion of the right SS (Fig. 3). The feeders were the OA and APA (Fig. 3B). To examine the relationship of the DAVF to the previously detected sinus thrombus, fusion three-

dimensional computer graphics (3DCG) images were reconstructed using GRID 1.1 software (Kompath Inc.). This application provides automatic image registration of multiple imaging studies by normalized mutual information.⁵ By integrating MRI and digital subtraction angiography, we found that the dilated feeder with neovascularization connected to the right SS along the previous thrombus site (Fig. 4C and D). A branch of the APA flowed into the SS at the site of the distal end of the previous thrombus, acting as the main feeder to the DAVF (Fig. 4). Because the DAVF was asymptomatic with no cortical venous reflux (CVR; i.e., Borden type I⁶ and Cognard type IIa⁷), the patient was managed conservatively with regular MRI follow-up. The angioarchitecture of the DAVF remained unchanged 27 months after its diagnosis.

Discussion

Observations

This is a unique report of a case in which we used 3DCG to demonstrate the chronological development of a DAVF after craniotomy for meningioma resection. The detailed architecture of a DAVF that developed secondary to postoperative sinus thrombosis could be clearly visualized by fusion 3DCG, integrating MRI and digital subtraction angiography. Through analysis of the fusion 3DCG images, we could infer the putative mechanism by which the DAVF developed after venous thrombosis.

DAVF after craniotomy is relatively rare, with few cases having been reported secondary to sinus thrombosis. In our literature review (Table 1), we found 25 cases of DAVF developing after craniotomy;^{8–26} 23 were directly related to the site of craniotomy and 2 appeared at different sites. There were 14 cases that developed after suboccipital craniotomy, 10 that represented postoperative stenosis or occlusion of the SS, and 2 that reflected postoperative thrombosis of the SS on imaging findings. The postoperative thrombosis of SS was detected 4⁹ and 6 months later,¹⁴ respectively. The median period to diagnosis of these DAVFs was 12 months (range, 4–240), and in those developing after suboccipital craniotomy, it was 24 months (range, 4–60). Postcraniotomy DAVF tends to develop slowly in patients with SS stenosis or occlusion after suboccipital craniotomy and generally appears at the site

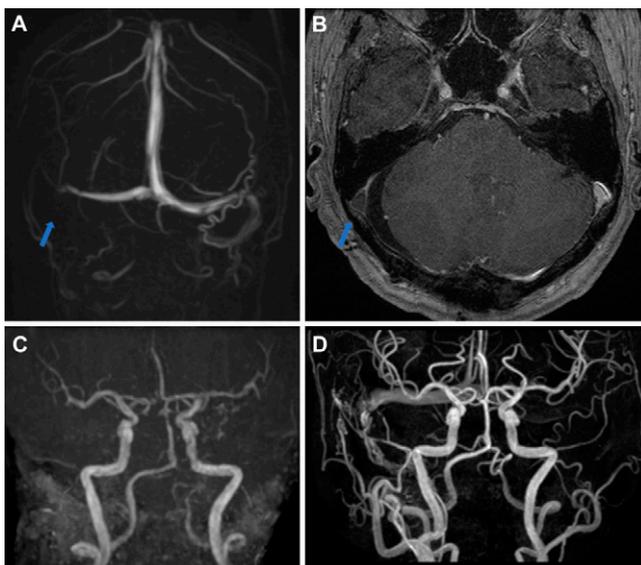


FIG. 2. Postoperative imaging. **A:** Three-dimensional gadolinium-enhanced MR venography on postoperative day 1. The right SS (*blue arrow*) was occluded. **B:** Axial gadolinium-enhanced MRI on postoperative day 1. Thrombus (*blue arrow*) was identified in the right sigmoid sinus. **C:** Three-dimensional time-of-flight MRA on postoperative day 1. No obvious vascular abnormalities had been observed before surgery. **D:** Three-dimensional time-of-flight MRA at 47 months postoperatively. The right transverse sinus-SS and dilated abnormal arteries are visualized.

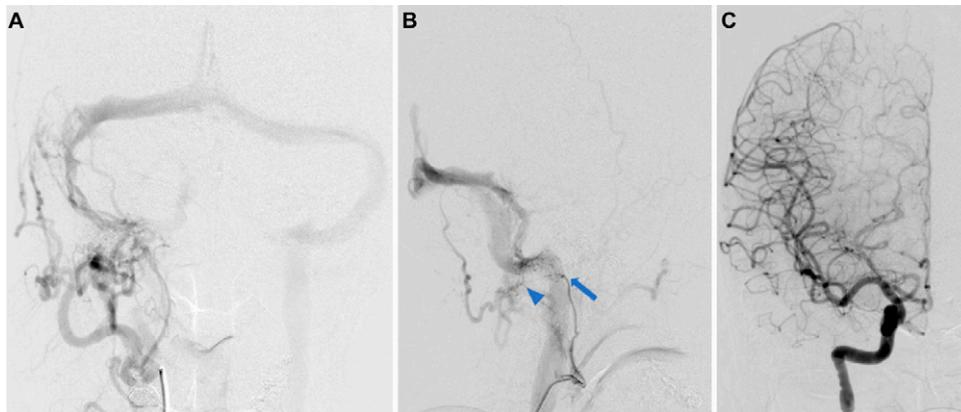


FIG. 3. Digital subtraction angiography of the dural arteriovenous fistula. **A:** Anteroposterior view of digital subtraction angiography of the right external carotid artery at 49 months postoperatively. A right SS DAVF with occlusion of the right SS was detected. **B:** Lateral view of digital subtraction angiography of the right external carotid artery at 49 months postoperatively. Microfeeders with neovascularization from the right APA (blue arrow) and the right dilated OA (blue arrowhead) were detected. **C:** Anteroposterior view of digital subtraction angiography of the right internal carotid artery at 49 months postoperatively. No vascular abnormalities were observed.

of craniotomy. These findings suggest that long-term surveillance for possible DAVFs may be necessary, especially in cases of SS stenosis or occlusion after suboccipital craniotomy.

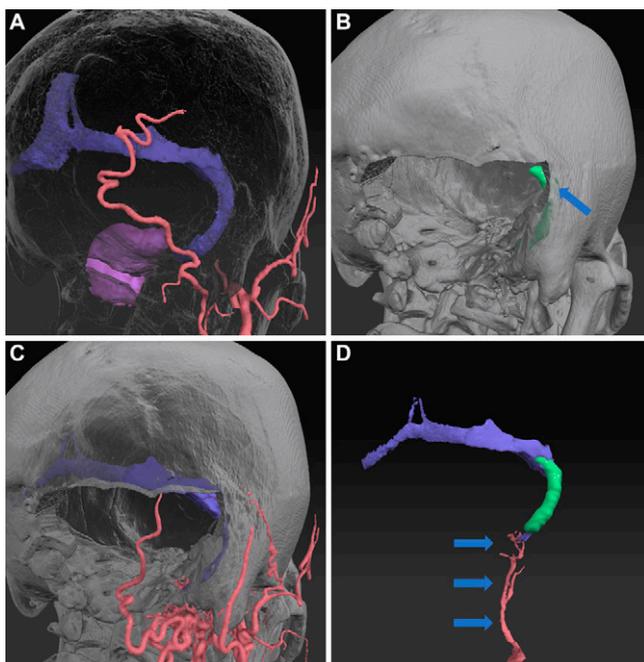


FIG. 4. Elucidation of the detailed architecture of the DAVF by fusion 3DCG. **A:** Fusion 3DCG before surgery. The positional relationship between the tumor (purple), SS (blue), and artery (red) is visualized. **B:** Fusion 3DCG on postoperative day 1. The thrombus (green) was manually reconstructed from loss of contrast in the sinus from T1-weighted MRI with contrast. Only the superior segment of the thrombus was at the superior corner of the craniotomy. **C:** Fusion 3DCG of the DAVF. The positional relationship among the SS, the artery, and craniotomy was revealed. The right dilated OA with neovascularization and partial recanalization of the right SS was observed. There were no obvious feeders passing through the site of craniotomy. **D:** Posterior view of fusion 3DCG without the skull bone. The main feeder was the right APA (blue arrows), which connected to the right SS at the distal point of the previous thrombus.

The involvement of sinus thrombosis in the development of DAVFs after craniotomy has been previously reported.²⁷ Venous hypertension is induced as a result of sinus stenosis or occlusion secondary to sinus thrombosis, which is a surgical complication that can occur particularly after suboccipital craniotomy.²⁸ Terada et al.²⁹ stated that venous hypertension can induce a DAVF. They estimated that increased venous pressure stimulates angiogenesis, resulting in direct connections to the sinus or vein and, ultimately, dural fistulas. Uranishi et al.³⁰ described how angiogenic growth factors, which are produced subsequent to sinus thrombosis and venous hypertension, may be implicated in the development of DAVF. Thus, dural venous sinus stenosis or thrombosis-induced secondary angiogenesis accompanied by venous hypertension may underlie the development of postoperative DAVF, but the precise pathogenesis remains unclear.

In our case, we visualized the detailed architecture of a DAVF that developed secondary to postoperative sinus thrombosis using fusion 3DCG. What is noteworthy about this approach is that it could clearly show how the DAVF developed secondary to postoperative sinus thrombosis. It is also remarkable that the fusion 3DCG demonstrated the positional relationship between the shunt point of the DAVF and the thrombus, showing how branches of the OA and APA extended with neovascularization to the SS along the previous thrombus site. These findings potentially corroborate the aforementioned hypothesis in which sinus thrombosis and venous hypertension are related to the development of DAVF.

Regarding intervention for delayed DAVF after craniotomy, successful management by either surgery or endovascular treatment has been reported (Table 1). Most symptomatic DAVFs after craniotomy in previous reports were treated. However, an asymptomatic case with spontaneous resolution²⁰ and a case with only mild tinnitus that was followed for 4 years with no change¹⁴ have also been reported. Observation is a reasonable option for asymptomatic DAVF without CVR.^{7,31} During follow-up of patients with DAVF, in addition to the presence of symptoms, the presence of CVR is another key factor in deciding the indication for treatment, which should be examined regularly. Lin et al.³² demonstrated how both

TABLE 1. Clinical characteristics of dural arteriovenous fistulas reported after craniotomy

Authors & Year	Age (yrs), Sex	Primary Pathology	Surgical Procedure	Sinus Stenosis/ Occlusion	Postoperative Sinus Thrombosis	Interval for DAVF Dev (mos postop)	Location of DAVF	Symptoms of DAVF
Nabors et al., 1987 ⁶	70, F	Rt trigeminal neuralgia	Rt SOC	NM	NM	4	Rt SS	Bruit
Sasaki et al., 1995 ⁹	60, M	Lt hemifacial spasm	Lt SOC	NM	NM	24	Lt SS	Bruit, tinnitus
Kim et al., 2014 ¹⁰	58, M	Rt trigeminal neuroma	Rt transpetrosal & transtentorial approach	Postoperative rt SS occlusion	Yes (rt SS)	23	Rt TS-SS	Bruit, dementia, gait disturbance
Yokoyama et al., 2019 ¹¹	49, M	Lt hemifacial spasm	Lt SOC	NM	NM	10	Lt TS-SS	ICH
	63, F	Lt CPA meningioma	Lt SOC	Postoperative lt SS occlusion	NM	6	Lt TS-SS	Bruit
	56, F	Rt CPA epidermoid tumor	Rt SOC & transpetrosal approach	Postoperative rt SS occlusion	NM	24	Rt TS-SS	Bruit
Sakaki et al., 1996 ¹²	59, F	Lt retromastoid meningioma	Lt SOC	Preoperative lt SS occlusion	NM	42	Lt TS-SS	ICH
	65, M	Rt jugular tubercle meningioma	Rt SOC	Rt SS was resected	NM	60	Rt TS-SS	HA, vomit, vertigo
	56, M	Rt hypoglossal neurinoma	Rt SOC	Preoperative rt SS occlusion	NM	54	Rt TS-SS	Cerebellar infarction
	31, F	Lt glomus jugulare tumor	Lt SOC	Lt SS was resected	NM	28	Lt TS-SS	Tinnitus
Xue et al., 2019 ¹³	50, F	Petroclival meningioma	Rt SOC	Postoperative rt SS stenosis	NM	24	Rt TS-SS	Tinnitus, audible behind
Yassari et al., 2002 ¹⁴	24, F	Pilocytic astrocytoma	Midline SOC	Postoperative lt SS occlusion	Yes (lt SS)	11	Lt TS-SS	Bruit, tinnitus
Higashida et al., 2015 ¹⁵	64, F	Cerebrospinal fluid otorrhea	Rt SOC	Postoperative rt SS occlusion	NM	48	Rt TS-SS	Tinnitus, dementia, gait disturbance
Sadahiro et al., 2014 ¹⁶	37, F	Brain stem cavernous hemangioma	Midline SOC	NM	NM	9	Lt inferior vermillion vein	NM
Dudeck et al., 2004 ¹⁷	16, M	Rt cerebellar DVA	Rt SOC	NM	NM	9	Rt TS-SS	Tinnitus, HA, blurred vision
Pabaney et al., 2016 ¹⁸	62, M	Epilepsy	Rt temporal craniotomy	NM	NM	240	Previous craniotomy site	SAH

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TABLE 1. Clinical characteristics of dural arteriovenous fistulas reported after craniotomy

Authors & Year	Age (yrs), Sex	Primary Pathology	Surgical Procedure	Sinus Stenosis/Occlusion	Postoperative Sinus Thrombosis	Interval for DAVF Dev (mos postop)	Location of DAVF	Symptoms of DAVF
Davie et al., 1967 ¹⁹	47, F	Lt sphenoid wing meningioma	Lt frontotemporal craniotomy	NM	NM	90	Previous craniotomy site	HA, loss of vision, proptosis
Peeters et al., 2020 ²⁰	NM	Moyamoya disease	Rt STA-MCA bypass	NM	NM	8	Previous craniotomy site	NM
Ugrinowski et al., 1989 ²¹	46, F	Rt falx meningioma	Rt parietal craniotomy	NM	NM	6	Far from previous craniotomy site	Bruit, tinnitus
Watanabe et al., 1984 ²²	59, F	Rt IC-PC AN	Rt frontotemporal craniotomy	NM	NM	4	Cavernous sinus	Tinnitus
Ding et al., 2016 ²³	57, F	Craniopharyngioma	Rt frontoparietotemporal craniotomy	NM	NM	10	Lt TS-SS	Bruit, tinnitus
Ahn et al., 2002 ²⁴	26, M	Rt ruptured AVM	NM	NM	NM	5	Previous craniotomy site	NM
Hashimoto et al., 1998 ²⁵	69, M	Lt ruptured AVM	Lt temporal craniotomy	NM	NM	12	Previous craniotomy site	NM
Diana et al., 2021 ²⁶	49, M	SAH due to ACOM AN	Bifrontal craniotomy	NM	NM	48	Anterior cranial fossa	ICH
	71, M	Rt AVM	Rt temporal craniotomy	NM	NM	4	Previous craniotomy site	Subacute subdural hematoma

ACOM = anterior communicating artery; AN = aneurysm; AVM = arteriovenous malformations; CPA = cerebellopontine angle; Dev = development; DVA = developmental venous anomaly; HA = headache; ICH = intracranial hemorrhage; IC-PC = internal carotid-posterior communicating artery; MCA = middle cerebral artery; NM = not mentioned; SAH = subarachnoid hemorrhage; SOC = suboccipital artery; STA = superficial temporal artery; TS = transverse sinus.

CTA and MRI/MRA have good diagnostic accuracy for detection of CVR in DAVF. Therefore, routine follow-up with cerebral angiography may not be mandatory given the associated risks. We are conservatively observing our patient using regular MRI/MRA. The lesion has remained stable to date.

Lessons

Our patient experienced a case of delayed postoperative DAVF secondary to SS thrombosis after removal of a large foramen magnum meningioma. Fusion 3DCG demonstrated that the feeders of the DAVF extended to the SS at the previous thrombus site. This finding implies that SS occlusion due to sinus thrombus is associated with the development of postoperative DAVF. There is a risk of delayed DAVF after craniotomy, and patients should be monitored carefully, especially in the case of SS stenosis or occlusion with sinus thrombosis after suboccipital craniotomy.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Miyawaki, Yajima. Acquisition of data: Miyawaki, Yajima, Kiyofuji, Kin. Analysis and interpretation of data: Miyawaki, Yajima, Kiyofuji. Drafting the article: Miyawaki, Yajima. Critically revising the article: Miyawaki, Koizumi, Kiyofuji, Hongo,

Segawa. Reviewed submitted version of manuscript: Miyawaki, Koizumi, Kiyofuji, Hongo, Segawa, Nakatomi, Saito. Approved the final version of the manuscript on behalf of all authors: Miyawaki. Administrative/technical/material support: Miyawaki. Study supervision: Miyawaki, Nakatomi.

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