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...
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 43 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...
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.

The involvement of sinus thrombosis in the development of DAVFs after craniotomy has been previously reported. 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. Terada et al. 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. 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 resolution and a case with only mild tinnitus that was followed for 4 years with no change have also been reported. Observation is a reasonable option for asymptomatic DAVF without CVR. 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. 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<sup>8</sup> | 70, F         | Rt trigeminal neuralgia  | Rt SOC                                    | NM                       | NM                            | 4                                 | Rt SS          | Bruit                           |
|                      |               |                          |                                            |                          |                               |                                   |                |                                 |
|                      | 60, M         | Lt hemifacial spasm      | Lt SOC                                    | NM                       | NM                            | 24                                | Lt SS          | Bruit, tinnitus                  |
| Sasaki et al., 1995<sup>9</sup> | 58, M         | Rt trigeminal neuroma     | Rt transpetrosal & transtentorial approach | Postoperative rt SS occlusion | Yes (rt SS)                  | 23                                | Lt TS-SS        | Bruit, dementia, gait disturbance|
|                      |               |                          |                                            |                          |                               |                                   |                |                                 |
| Kim et al., 2014<sup>10</sup> | 49, M         | Lt hemifacial spasm      | Lt SOC                                    | NM                       | NM                            | 10                                | Lt TS-SS        | ICH                             |
| Yokoyama et al., 2019<sup>11</sup> | 63, F         | Lt CPA meningioma        | Lt SOC                                    | Postoperative lt SS occlusion | NM                          | 6                                 | Lt TS-SS        | Bruit                           |
|                      |               |                          |                                            |                          |                               |                                   |                |                                 |
|                      | 56, F         | Lt CPA epidermoid tumor  | Rt SOC & transpetrosal approach           | Postoperative rt SS occlusion | NM                          | 24                                | Rt TS-SS        | Bruit                           |
| Sakaki et al., 1996<sup>12</sup> | 59, F         | Lt retromastoid meningioma | Lt SOC                                  | Preoperative lt SS occlusion | NM                          | 42                                | Lt TS-SS        | ICH                             |
|                      |               |                          |                                            |                          |                               |                                   |                |                                 |
|                      | 65, M         | Rt jugular tuberde 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<sup>13</sup> | 50, F         | Petroclival meningioma   | Rt SOC                                    | Postoperative rt SS stenosis | NM                            | 24                                | Rt TS-SS        | Tinnitus, audible behind         |
|                      |               |                          |                                            |                          |                               |                                   |                |                                 |
|                      | 24, F         | Pilocytic astrocytoma    | Midline SOC                               | Postoperative lt SS occlusion | Yes (lt SS)                  | 11                                | Lt TS-SS        | Bruit, tinnitus                  |
| Higashida et al., 2015<sup>15</sup> | 64, F         | Cerebrospinal fluid otorhea | Rt SOC                                    | Postoperative rt SS occlusion | NM                           | 48                                | Rt TS-SS        | Tinnitus, dementia, gait disturbance |
| Sadahiro et al., 2014<sup>16</sup> | 37, F         | Brain stem cavernous hemangioma | Midline SOC                           | NM                       | NM                            | 9                                 | Lt inferior vermian vein |                               |
| Dudeck et al., 2004<sup>17</sup> | 16, M         | Rt cerebellar DVA        | Rt SOC                                    | NM                       | NM                            | 9                                 | Rt TS-SS        | Tinnitus, HA, blurred vision     |
| Pabaney et al., 2016<sup>18</sup> | 62, M         | Epilepsy                  | Rt temporal craniotomy                   | NM                       | NM                            | 240                               | Previous craniotomy site        | SAH              |

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| 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<sup>19</sup> | 47, F | Lt sphenoid wing meningioma | Lt frontotemporal craniotomy | NM | NM | 90 | Previous craniotomy site | HA, loss of vision, proptosis |
| Peeters et al., 2020<sup>20</sup> | NM | Moyamoya disease | Rt STA-MCA bypass | NM | NM | 8 | Previous craniotomy site | NM |
| Ugrinovski et al., 1989<sup>21</sup> | 46, F | Rt falx meningioma | Rt parietal craniotomy | NM | NM | 6 | Far from previous craniotomy site | Bruit, tinnitus |
| Watanabe et al., 1984<sup>22</sup> | 59, F | Rt IC-PC AN | Rt frontotemporal craniotomy | NM | NM | 4 | Cavernous sinus | Tinnitus |
| Davie et al., 1967<sup>19</sup> | 57, F | Craniopharyngioma | Lt frontoparietotemporal craniotomy | NM | NM | 10 | Lt TS-SS | Bruit, tinnitus |
| Ding et al., 2016<sup>23</sup> | 26, M | Rt ruptured AVM | NM | NM | NM | 5 | Previous craniotomy site | NM |
| Ahn et al., 2002<sup>24</sup> | 69, M | Lt ruptured AVM | Lt temporal craniotomy | NM | NM | 12 | Previous craniotomy site | NM |
| Hashimoto et al., 1998<sup>25</sup> | 49, M | SAH due to ACOM AN | Bilfrontal craniotomy | NM | NM | 48 | Anterior cranial fossa | ICH |
| Diana et al., 2021<sup>26</sup> | 71, M | Rt AVM | Lt 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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