Intracranial Dural Arteriovenous Fistulas with Pial Arterial Supply

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BACKGROUND: Pial arterial supplies are sometimes found in patients with dural arteriovenous fistulas (DAVFs), though their characteristics have rarely been clarified.
OBJECTIVE: To investigate the characteristics of pial arterial supplies in DAVFs and to discuss their pathophysiology and treatment.
METHODS: Two hundred four consecutive patients with intracranial DAVFs over 11 yr were retrospectively reviewed. Clinical factors and radiological findings, including the presence of pial arterial supplies, were evaluated. Supply from a pial artery was classified into 2 categories: dilated pre-existing dural branches of pial arteries, and a "pure" pial supply.
RESULTS: Twenty-three of 204 patients (11.3%) showed an additional pial arterial supply. Multivariate analysis identified 3 independent predictors of a pial arterial supply: younger age ($P < .0005$), DAVF within the tentorium ($P = .0162$), and presence of venous dilatation ($P = .0001$). A dilated pre-existing dural branch of a pial artery was identified in 17 patients, while 8 had a pure pial supply. Of these 23 patients, 17 underwent interventional therapy. No postoperative intracranial hemorrhage or infarction occurred in patients with pial arterial supplies.
CONCLUSION: An additional pial supply is not uncommon in DAVFs and may be explained by a rich physiological pial arterial supply to the dura mater from the posterior circulation, while potential angiogenesis due to venous hypertension remains speculative. Prior to interventional treatment for DAVFs, recognition of a pial arterial supply to the DAVF might influence the treatment strategy and could help avoid inadvertent retrograde embolization of brain supplying vessels through the pial network.

KEY WORDS: Dural arteriovenous fistula, Pial arterial supply, Dural branch, Pial supply, Pure pial supply, Sprouting angiogenesis

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Cranial dural arteriovenous fistulas (DAVFs) are abnormal shunts from arteries to veins and/or venous sinuses located in the dura mater. It has been recognized that the blood supply to these shunts can, albeit rarely, also be provided by pial arteries.[1-5] A pial arterial supply to DAVFs has been reported to develop after cerebral venous sinus thrombosis; therefore, an anastomosis between pial arteries and dural arteries has been considered an acquired lesion.[6,7] A higher rate of procedure-related complications was reported in the treatment of DAVFs with a pial arterial supply than in those without an additional pial arterial supply.[4] Identification of the characteristics of the pial arterial supply in DAVFs, including their frequency, locations, and morphological features, may help clarify the mechanisms of development of a pial arterial supply. Furthermore, awareness of these vessels may help avoid complications related to the reflux of embolic agents into pial vessels. To the best of our knowledge, no previous paper has reported a consecutive series of patients with

ABBREVIATIONS: **ACF,** anterior cranial fossa; **ADS,** artery of Davidoff and Schechter; **AICA,** anterior inferior cerebellar artery; **CS,** cavernous sinus; **CT,** computed tomography; **DAVF,** dural arteriovenous fistula; **FM,** foramen magnum; **HIF,** hypoxia-inducible factor; **MCF,** middle cranial fossa; **MR,** magnetic resonance; **OR,** odds ratio; **PCA,** posterior cerebral artery; **PICA,** posterior inferior cerebellar artery; **SCA,** superior cerebellar artery; **SSS,** superior sagittal sinus; **TSS,** transverse sigmoid sinus; **VEGF,** vascular endothelial growth factor

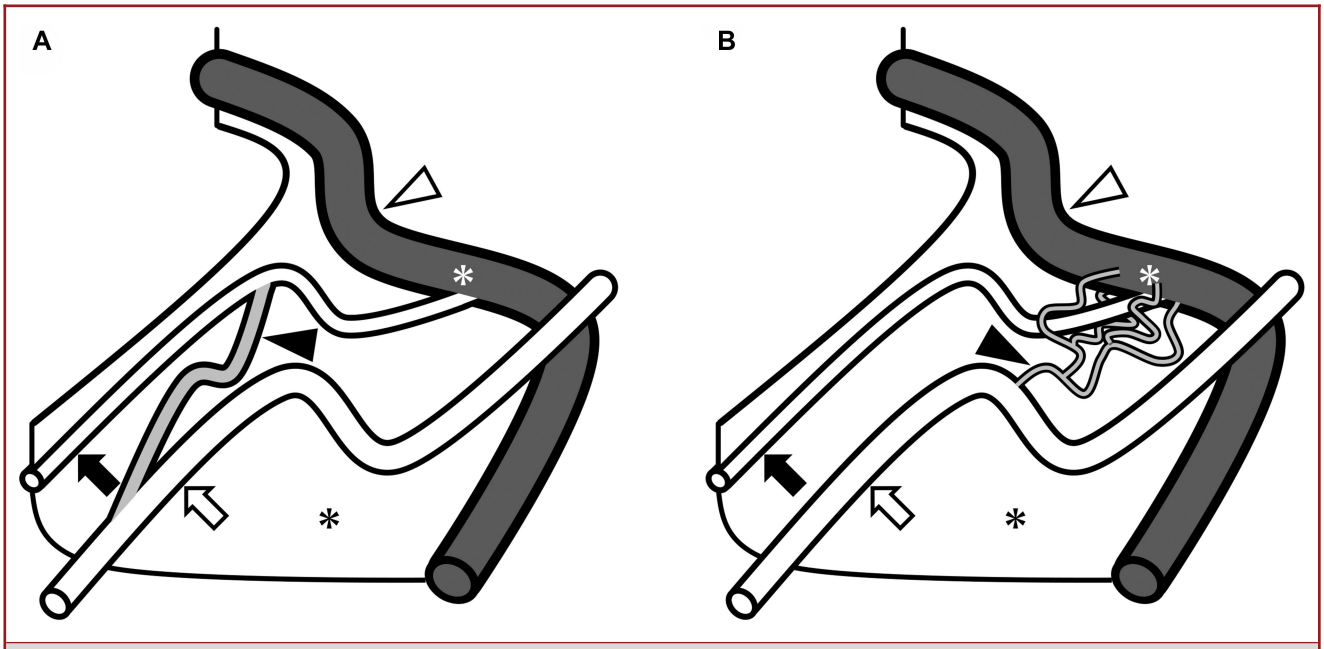

FIGURE 1. *The schema of the classification of pial arterial supplies in DAVFs. A meningeal artery (black arrow) runs on the outer surface of the dura (black asterisk) and anastomoses with a sinus (white arrow head) at a shunt point (white asterisk). Pial arterial supplies (black arrow head) arising from a pial artery (white arrow) are classified into 2 categories.* **A**, *Dilated pre-existing dural branch of a pial artery is a morphologically linear vessel (black arrow head).* **B**, *A pure pial supply is a morphologically tortuous and ramified vessel.*

DAVFs in which the characteristics of the pial arterial supply were reviewed.

In the present study, the characteristics of DAVFs with pial arterial supplies were examined and compared to DAVFs without pial arterial supplies. An attempt was made to classify the pial arterial supplies based on anatomic and morphological criteria into 2 categories. Hypotheses for the pathophysiology of the development of pial arterial supplies and treatment strategies for DAVFs with pial arterial supplies are discussed.

METHODS

Patient Population

Using a prospectively collected, dedicated neurovascular database, consecutive patients with intracranial DAVFs who were managed at our hospital between January 2006 and December 2016 were retrospectively investigated. Patients who did not undergo treatment were included. Patients with a direct carotid-cavernous fistula or spinal DAVF were excluded. Patients with DAVFs developing after craniotomy were also excluded. Clinical factors, including age, sex, and clinical symptoms at onset, were investigated by review of the patients' charts. Approval for this study was obtained from the institutional review board at our institution.

Image Acquisition and Imaging Analysis/Interpretation

Radiological findings, including angiography, of DAVFs were categorized according to both the Borden and Cognard classifications.[8,9] According to the cerebral angiography findings, locations of DAVFs were classified into cavernous sinus (CS), transverse sigmoid sinus (TSS), torcular, tentorium, anterior cranial fossa (ACF), middle cranial fossa (MCF), foramen magnum (FM; including anterior condylar confluent), convexity, superior sagittal sinus (SSS), and falx cerebri. A DAVF located in 2 or more locations was defined as an "extensive" fistula spreading to multiple locations continuously or as "multiple" fistulas when the lesions were separate. On angiography, the presence of venous dilatation, varices, and venous congestion was evaluated. Venous dilatation was defined as greater dilatation of veins than usual, and a varix was defined as a venous dilatation with an axial diameter more than twice as large as that of veins proximal and distal to the varix. Venous congestion was defined as a pseudophlebitic pattern, as reported by Willinsky et al.[10] Tentorial DAVFs were divided into 4 subtypes according to Lawton's classification.[3] Basically, the presence of a pial arterial supply to a DAVF was evaluated using conventional angiography. When the angioarchitecture of the DAVF, including the suspected existence of a pial arterial supply, was not clearly demonstrated by conventional angiography, superselective angiography was performed. A pial arterial supply was defined as a blood supply from a pial artery to fistulas located within the dura mater.

Pial arterial supplies were classified into 2 categories based on morphological and anatomic findings (Figure 1). One was characterized by linear vessels that were dilated "physiological" pre-existing dural branches of pial arteries.[1] This type of pial arterial supply was categorized as dilated dural branches of pial arteries. The second category consisted of tortuous and ramified vessels that were present in locations other than the known physiological connections between pial arteries and dura mater. This type was referred to as a "pure" pial supply. Angiograms were evaluated by a neuroradiologist and a neurosurgeon with more than 15 yr of experience

in their respective specialties. After independent review, a consensus reading was performed to resolve any differences in interpretation and to determine the type of pial arterial supply.

On preoperative computed tomography (CT) scans and/or magnetic resonance (MR) images, DAVF-induced abnormal findings, including cerebral edema, intracranial hemorrhage, and cerebral infarction, were evaluated. On CT scans and/or MR images taken within a few days after surgery, abnormal findings, including intracranial hemorrhage, were evaluated. In treated patients, DAVF obliteration was verified on delayed angiography or time-resolved MRA (typically 6-12 mo following the initial procedure).

Treatment

Patients with Borden type I DAVFs were typically managed conservatively with radiological and clinical follow-up. Patients with DAVFs of Borden type II or III underwent transarterial and/or transvenous interventional treatment, in which coils, N-butyl cyanoacrylate (Histoacryl, B. Braun, Melsungen, Germany), and/or ethylene vinyl alcohol copolymer (Onyx; ev3, Medtronic Inc, Dublin, Ireland), or a combination thereof was used through either transarterial or transvenous approaches, depending on the location and angioarchitecture of the lesion. When a residual type II or III fistula was observed on follow-up angiography, repeat interventional therapy, craniotomy, and/or gamma knife therapy was performed to ensure obliteration.

Statistical Analysis

In univariate analyses, chi-squared analysis was used for categorical variables and Student's *t*-test or the Mann–Whitney U-test for continuous variables. In multivariate analysis, logistic regression analysis was employed. We used variables, in which *P* values were less than .1 level in the univariate analyses, to perform the multivariate analysis. In the multivariate analysis, the variables were reduced by successive exclusion of the least significant variable from the model, and the final model contained only significant variables at $P < .05$ level. The variables with $P < .05$ were regarded as statistically significant predictors. Numerical data are expressed as means ±SD (standard deviation). All statistical analyses were conducted using JMP 10 (SAS Institute Inc, Cary, North Carolina).

RESULTS

Between January 2006 and December 2016, 204 patients with intracranial DAVFs were managed in our institution. Their mean age was 59.6 yr; 98 were male. Fistulas were located in the TSS in 72 patients, CS in 46, tentorium in 38, FM in 11, torcula in 7, along the convexity in 7, ACF in 6, falx cerebri in 4, SSS in 4, and MCF in 2, with extensive fistulas in 4, and multiple fistulas in 3 patients. On cross-sectional imaging, cerebral edema was observed in 30 patients (14.7%), and intracranial hemorrhage was seen in 36 (17.6%). Three patients (1.5%) showed cerebral infarction. Conventional 6-vessel cerebral angiography demonstrated venous dilatation, varices, and venous congestion in 88 (43.1%), 66 (32.4%), and 56 patients (27.5%), respectively.

Detection and Analysis of DAVFs with a Pial Arterial Supply

A pial arterial supply was observed in 23 of the 204 patients (11.3%). Table 1 shows the clinical characteristics of these 23 patients. The patients' mean age was 53.2 yr. Initial clinical presentations were pulsatile tinnitus in 7 patients, headache in 6, and intracranial hemorrhage in 5 (subarachnoid hemorrhage in 3 patients and intracerebral hemorrhage in 2). In 4 patients, the DAVFs were found incidentally. In 1 patient, the DAVF developed after previous sinus thrombosis. The DAVFs were located in the tentorium, TSS, SSS, torcular, and FM. An additional pial arterial supply was seen in 12 of 38 patients with tentorial DAVFs (31.6%), 8 of 72 patients with a TSS DAVF (11.1%), 1 of 4 patients with an SSS DAVF (25.0%), 1 of 7 patients with a torcular DAVF (14.3%), and 1 of 11 patients with an FM DAVF (9.1%).

Table 2 shows a comparison of DAVF patients with and without a pial arterial supply. Clinical factors used for multivariate analysis were patient age and sex, location of the DAVF, Borden and Cognard classification of DAVF, brain edema, intracranial hemorrhage, cerebral infarction, venous dilatation, varices, and venous congestion. Multivariate analysis identified 3 independent predictors of a pial arterial supply: age ($P = .0005$; odds ratio [OR]: 0.93), location of the DAVF at the tentorium ($P = .0162$; OR: 3.60), and venous dilatation on angiograms ($P = .0001$; OR: 10.72). With regard to the subtypes of the 38 tentorial DAVFs, a pial arterial supply was observed in 5 of 16 patients with an SPS DAVF (31.2%), 3 of 5 patients with a Galenic DAVF (60.0%), and 4 of 13 patients with an SS DAVF (30.8).

Classification of Pial Arterial Supplies to DAVFs

Pial arteries supplying DAVFs originated from the following arteries: the superior cerebellar artery (SCA) in 10 patients; the posterior cerebral artery (PCA) in 9; the anterior inferior cerebellar artery (AICA) in 4; the posterior inferior cerebellar artery (PICA) in 4; the anterior cerebral artery (ACA) in 2; and the middle cerebral artery (MCA) in 1. To clarify the angioarchitecture of DAVFs including the pial arterial supplies, superselective (microcatheter) angiography was performed following conventional angiography in 2 patients with pial arterial supplies from the SCA and in 1 patient with a pial arterial supply from the PCA. Of the 23 patients with pial arterial supplies, 17 showed 1 or more dilated dural branches of the pial artery, and 8 patients showed a pure pial supply. Among them, 2 patients showed both dilated dural branches and a pure pial supply. Table 3 shows the characteristics of each pial arterial supply. In 17 patients with a dilated pre-existing dural branch of a pial artery, 11 patients (64.7%) had tentorial DAVFs. On the other hand, 6 of 8 patients with a pure pial supply (75%) had TSS DAVFs. The mean age of patients showing a pure pial supply was 47.1 yr, which was younger than that of patients with a dilated dural branch of a pial artery (56.4 yr), but this difference was not significant.

TABLE 1. Details of Patients With Dural Arteriovenous Fistulas With Pial Arterial Supplies

No.	Age (yr)/sex	Location (subtype)	Initial clinical presentation	Borden/ Cognard	Pial arterial supply: Dilated dural branch	Pial arterial supply: Pure pial supply	Treatment	Final DAVF status
1	45/M	SSS	Tinnitus, headache	II/II a + b		ACA	Observation	No change
2	50/F	Tentorium (SPS)	Incidentally	III/IV		SCA	γ knife, TAE (Onyx), Craniotomy	Reduction
3	36/M	Tentorium (SPS)	Tinnitus	III/IV	AICA		TAE (NBCA), Craniotomy	Obliterated
4	48/M	Tentorium (SPS)	Hydrocephalus	III/IV	SCA		TAE(Onyx)	Obliterated
5	52/M	Tentorium (SPS)	Subarachnoid hemorrhage	III/IV	AICA		TAE (NBCA)	Obliterated
6	58/M	Tentorium (SPS)	Headache	III/IV	AICA		Observation	Unknown
7	67/M	Tentorium (SS)	Dizziness	III/IV	SCA		TAE (Onyx)	Obliterated
8	53/F	Tentorium (SS)	Subarachnoid hemorrhage	III/IV	PCA, PICA		TAE (ONYX)	Obliterated
9	70/M	Tentorium (SS)	Gait and memory disturbance	III/III	SCA		Craniotomy	Obliterated
10	64/M	Tentorium (SS)	Incidentally	III/IV	SCA		γ knife	Unknown
11	64/F	Tentorium (Galenic)	Incidentally	III/IV	PCA, ACA		TAE (Onyx), γ knife	Reduction
12	67/M	Tentorium (Galenic)	Headache	III/IV	PCA, SCA		γ knife	Unknown
13	31/M	Tentorium (Galenic)	Headache	III/IV	PCA, SCA		TAE (Onyx), TVE (coil, Onyx)	Obliterated
14	59/F	TSS	ICH	II/II a + b		MCA	γ knife	No change
15	58/F	TSS	Tinnitus	III/IV		PCA	TAE (Onyx)	Obliterated
16	23/M	TSS	Incidentally, Post sinus thrombosis	II/II a + b		PCA	TVE (coil)	Obliterated
17	41/F	TSS	Tinnitus, Headache	I/II a	SCA		TAE (NBCA)	Reduction
18	67/F	TSS	Tinnitus	III/III	PICA	PCA	TVE (coil), γ knife	Reduction
19	31/F	TSS	Tinnitus, Headache	II/II a + b		PCA	TVE (coil), TAE (NBCA)	Obliterated
20	44/F	TSS	Tinnitus	II/II a + b	SCA	PCA	TVE (coil), TAE (NBCA)	Obliterated
21	57/F	TSS	ICH	III/IV	SCA, AICA		TAE (NBCA)	Obliterated
22	71/M	Torcular	Memory disturbance	II/II a + b	PICA		TVE (coil)	Obliterated
23	68/F	FM	Subarachnoid hemorrhage	III/IV	PICA		TAE (Onyx)	Obliterated

SSS, superior sagittal sinus; TTS, transverse sigmoid sinus; SPS, superior petrosal sinus; SS, straight sinus; FM, foramen magnum; ICH, intracerebral hemorrhage; ACA, anterior cerebral artery; MCA, middle cerebral artery; PCA, posterior cerebral artery; SCA, superior cerebellar artery; AICA, anterior inferior cerebellar artery; PICA, posterior inferior cerebellar artery; TAE, transarterial embolization; TVE, transvenous embolization; NBCA, N-butyl cyanoacrylate

Treatment and Postoperative Course

In this series of 204 patients, interventional treatment, direct surgery, gamma knife surgery, and conservative management were performed in 127 (62.3%), 25 (12.3%), 10 (4.9%), and 51 (25%) patients, respectively. In the conservatively managed group, 9 patients proceeded to obliteration, and, following intervention, complete obliteration (to date) of DAVF was achieved in 121 of 153 patients, with conversion to a "benign" (Cognard or Borden Type I) DAVF in 17; in 8 patients, treatment is ongoing, 3 patients refused further management, and 2 patients were lost to follow-up or treatment was not continued (related to age in one and significant comorbidities in the other). The conservatively managed patients were followed clinically.

Interventional treatment was performed in 17 of 23 patients with DAVFs associated with a pial arterial supply. Embolization via a pial artery was performed in 1 patient with a dilated

TABLE 2. Comparison Between Patients With And Without a Pial Arterial Supply in 204 Cases of Intracranial Dural Arteriovenous Fistulas

Variable	With pial arterial supply (n = 23)	Without pial arterial supply (n = 181)	*P* value univariate analysis	Multivariate analysis	
				OR (95% CI)	*P* value
Age (yr)	53.2 ± 13.9	60.4 ± 12.3	.0259	0.93 (0.89-0.97)	.0005
Female	11 (47.8%)	95 (52.5%)	.6735		
Location of DAVF					
Transverse sigmoid sinus	8 (38.1%)	64 (35.0%)	.7767		
Cavernous sinus	0 (0%)	46 (25.1%)	.0090		
Tentorium	12 (57.1%)	26 (14.2%)	<.0001	3.60 (1.27-10.58)	.0162
Foramen Magnum	1 (4.4%)	10 (5.5)	.8139		
Torcula	1 (4.4%)	6 (3.3%)	.7977		
Convexity	0 (0%)	7 (3.8%)	.3617		
Anterior cranial fossa	0 (0%)	6 (3.3%)	.3996		
Extensive fistula	0 (0%)	4 (2.2%)	.4938		
Falx cerebri	0 (0%)	4 (2.2%)	.4938		
Superior sagittal sinus	1 (4.4%)	3 (1.6%)	.3283		
Multiple fistulas	0 (0%)	3 (1.6%)	.5545		
Middle cranial fossa	0 (0%)	2 (1.1%)	.6302		
Classification					
Borden	2.65 ± 0.57	2.02 ± 0.93	<.0001		
Cognard	3.35 ± 0.88	2.52 ± 1.14	.0003		
Medical imaging					
Brain edema	4 (18.1%)	26 (16.1%)	.7995		
Intracranial hemorrhage	3 (13.0%)	31(18.3%)	.5322		
Cerebral infarction	1 (4.4%)	2 (1.2%)	.2609		
Venous dilatation	20 (87.0%)	68 (41.0%)	<.0001	10.72 (2.97-52.92)	.0001
Varix	17 (73.9%)	49 (29.5%)	<.0001		
Venous congestion	9 (40.9%)	47 (28.5%)	.2320		

DAVF, dural arteriovenous fistula.

TABLE 3. Characteristics of Dural Arteriovenous Fistulas With Pial Arterial Supplies Within Each Classification

	Classification of pial arterial supplies	
	Dilated dural branch (n = 17)	Pure pial supply (n = 8)
Age (mean ± SD), yr	56.4 ± 3.1	47.1 ± 14.7
Supplying arteries	ACA: olfactory branches and pericallosal branches (n = 1)	ACA (n = 1)
	PCA: artery of Davidoff and Schechter (n = 4)	MCA (n = 1)
	SCA: medial dural tentorial branch (n = 9)	PCA (n = 5)
	AICA: subarcuate artery (n = 4)	SCA (n = 1)
	PICA: posterior meningeal artery or artery of the falx cerebelli (n = 4)	
Morphological feature	Linear vessel	Tortuous and ramified vessel
Location of DAVF	Tentorium (n = 11)	Tentorium (SPS) (n = 1)
	(SPS [n = 4], SS [n = 4], Galen [n = 3])	TSS (n = 6)
	TSS (n = 4)	SSS (n = 1)
	Torcular (n = 1)	
	FM (n = 1)	

DAVF, dural arteriovenous fistula; ACA, anterior cerebral artery; MCA, middle cerebral artery; PCA, posterior cerebral artery; SCA, superior cerebellar artery; AICA, anterior inferior cerebellar artery; PICA, posterior inferior cerebellar artery; SSS, superior sagittal sinus; TTS, transverse sigmoid sinus; SPS, superior petrosal sinus; SS, straight sinus; FM, foramen magnum

Five patients showed multiple dilated dural branches supplied by multiple cerebral arteries.

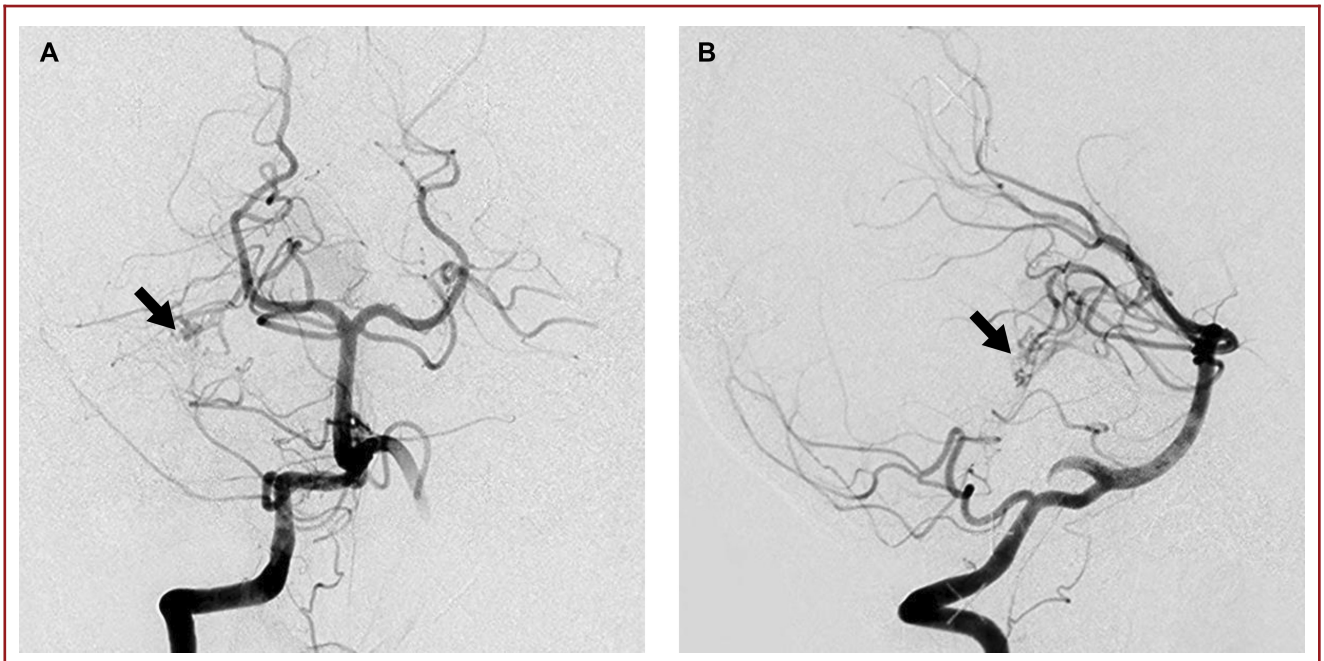

FIGURE 2. *A right vertebral angiogram of case 2 showing a superior petrosal sinus DAVF. An arrow indicates the pure pial supply from the SCA (***A***, anteroposterior [AP] view;* **B***, lateral view).*

pre-existing dural branch and 1 patient with a pure pial arterial supply. In 8 of the 17 patients, residual fistulas were observed on angiograms immediately after treatment. In 5 patients, the pial arterial supply was also still observed. In 1 patient, the fistula subsequently occluded. In 5 additional patients with residual DAVFs, additional treatments resulted in obliteration; thus, complete obliteration was obtained in 14 cases. In the remaining 3, flow reduction or conversion to a benign fistula was achieved.

Intracranial hemorrhage was not seen on CT scans immediately after treatment. Among all 204 patients with DAVFs, 1 patient without a pial arterial supply showed intracranial hemorrhage on a CT scan immediately after direct surgery. In the present series, several patients showed retrograde propagation of the liquid embolic material towards the dural artery connecting to the pial artery, but pushing of the embolic agent was stopped, and no patient showed ischemic complications caused by iatrogenic retrograde embolization.

On longer term follow-up (between 6 mo and 1 yr after treatment), patients with confirmed angiographic occlusion remained occluded. New hemorrhagic events occurred in 1 case, with the DAVF located in the TSS with a varix and without a pial arterial supply. This event was observed 3 mo after interventional therapy.

Illustrative Cases

Case 2

A 50-yr-old woman had an incidental SPS DAVF. Angiography showed a pial arterial supply from the right SCA (Figure 2). Although there is a known dural supply arising from the SCA, this supply was classified as "pure pial" given the course of the artery and the location of the supply. The patient had previously undergone gamma knife surgery. Embolization of the DAVF via the MMA followed by open surgery led to flow reduction of the DAVF.

Case 7

A 67-yr-old man presented with dizziness. Angiography showed an SS DAVF with a pial arterial supply from the left SCA (Figure 3). The pial arterial supply was classified as a dilated pre-existing dural branch. Embolization via the MMA resulted in complete obliteration of the DAVF.

Case 11

A 64-yr-old woman had an incidental Galenic DAVF. Angiography showed the pial arterial supply from the bilateral PCAs and ACAs (Figure 4). The pial arterial supply was through a dilated pre-existing dural branch. Embolization of the DAVF via the

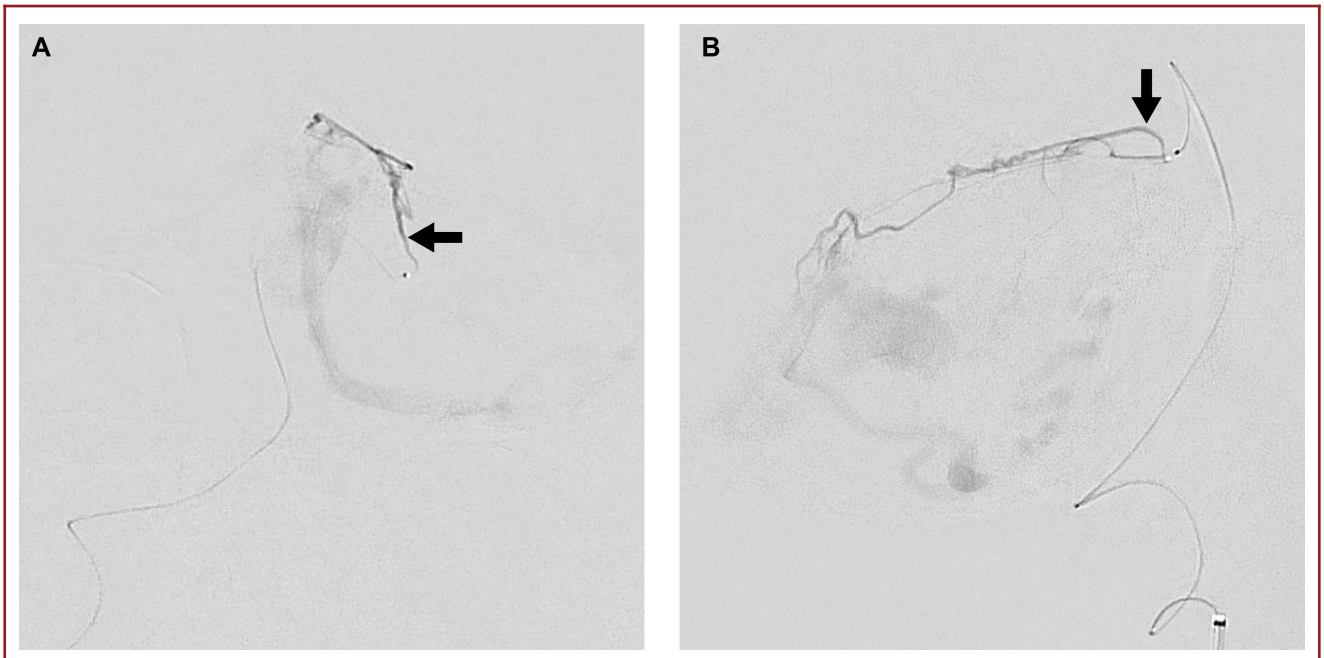

FIGURE 3. *A selective angiogram of case 7 with a straight sinus DAVF showing the medial dural-tentorial branch (arrow) from the SCA (**A**, AP view; **B**, lateral view).*

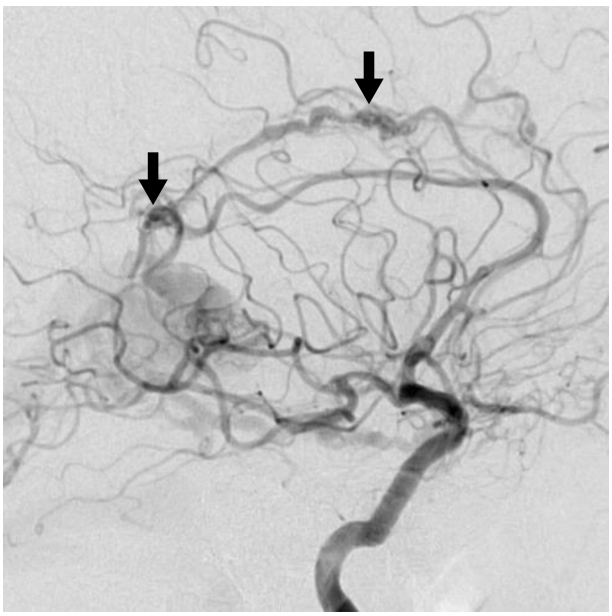

FIGURE 4. *A left internal carotid angiogram of case 11 showing a Galenic DAVF (lateral view). A dilated dural branch of a pial artery (arrow) arises from the pericallosal and callosomarginal arteries.*

MMA, OA, and superficial temporal artery, followed by gamma knife treatment, resulted in flow reduction of the DAVF.

Case 13

A 31-yr-old woman had a DAVF in the Galenic sinus and presented with headache. Angiography showed a pial arterial supply from the right PCA and the left SCA related to a dilated pre-existing dural branch (Figure 5). After transarterial embolization through the MMA, complete occlusion of the DAVF was achieved by transvenous embolization.

Case 16

A 23-yr-old man showed a TSS DAVF after sinus thrombosis. Angiography showed a pial arterial supply from the right PCA (Figure 6). The pial arterial supply was classified as a "pure" pial supply. Transvenous embolization resulted in complete obliteration of the DAVF.

DISCUSSION

In this study, an additional pial arterial supply was observed in more than 10% of patients harboring a DAVF (11.3%; 23 of 204). The pial arterial supply could be classified into 2 categories: dilated pre-existing dural branches between pial arteries and the dura mater; and a pure pial supply, the pathophysiological origin of which remains unknown, with angiogenesis being a yet to be proven hypothesis.

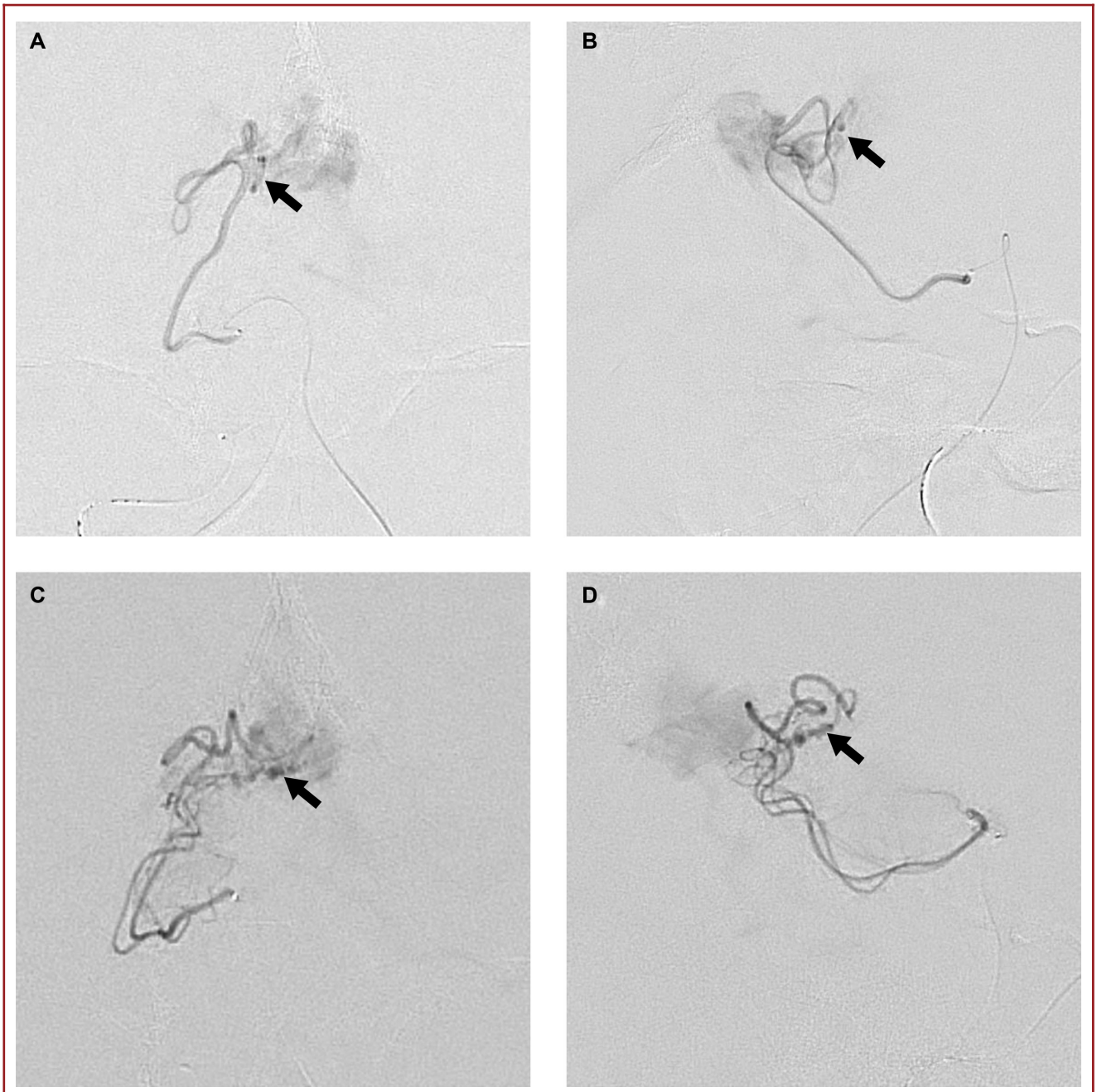

FIGURE 5. *A selective angiogram of case 13 showing a Galenic DAVF supplied from the PCA. The ADS is demonstrated in* **A** *(AP view) and* **B** *(lateral view). A circumferential branch from the PCA to the dura mater is shown in* **C** *(AP view) and* **D** *(lateral view). Both of them are regarded as dilated pre-existing dural branches of pial arteries.*

Anatomy of Dilated Physiological Dural Branches of Pial Arteries Supplying DAVFs

In the present series, 15 patients showed pial arterial supplies related to dilated dural branches of pial arteries. Several previous studies have reported that various physiological dural branches from pial arteries are present in normal subjects.[1,11,12] Dural branches that originate from the ACA have been reported to have their origin at several locations.[1] The olfactory branch of the orbitofrontal branch of the ACA shows anastomoses with the

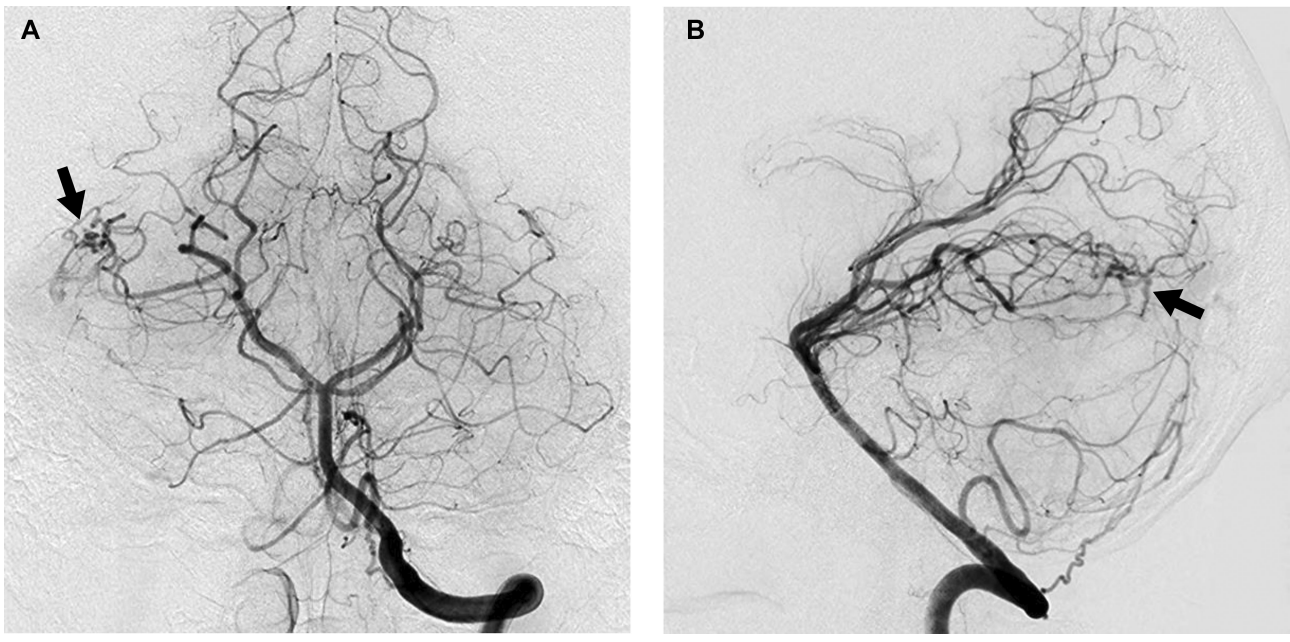

FIGURE 6. *A left vertebral angiogram of case 16 showing a transverse sigmoid sinus DAVF. An arrow indicates the pure pial supply from the PCA (**A**, AP view; **B**, lateral view).*

ethmoidal arteries arising from the ophthalmic artery. In support of this potential route of supply is a case of an ACF DAVF supplied from a persistent primitive olfactory artery.[13] Another dural branch that originates from a distal part of the ACA is a connection between the pericallosal artery and the falx cerebri. This type of distal branch was frequently observed in cases of SSS DAVFs.[1] In case 11 of the present series, the dilated distal branch of the ACA was shown to supply Galenic DAVFs (Figure 4).

A well-known dural branch of the PCA, also called the artery of Davidoff and Schechter (ADS),[14,15] arises from the P2 segment of the PCA and supplies the tentorial incisura. It is observed in 10% of normal autopsy cases.[11] Blood supply from the ADS has been reported in cases of DAVFs within the parieto-occipital region, meningiomas of the posterior falx cerebri, and cerebellar hemangioblastomas.[1,16,17] In case 13 of the present study, pial arterial supplies from both the ADS and circumferential branches of the PCA were demonstrated by selective angiography (Figure 5).

Physiological dural branches arising from the SCA to the tentorial incisura have been reported in angiographic studies, cadaver studies, and intraoperative findings.[1,2,12,18] These dural branches were demonstrated in cases 7, 15, and 20 of this series (Figure 3). A recent study reported that a pial arterial supply from the PCA and SCA was observed in 6 of 7 patients with Galenic DAVFs (86%).[3]

The subarcuate artery, which is a dural branch of the AICA, arises distal to the meatal loop of the AICA, runs close to the dura mater, and then enters the subarcuate canal. A cadaver dissection study reported that the subarcuate artery arose from the AICA in 80%, the accessory AICA in 17%, and the PICA in 3% of cases.[19] Cases 3, 5, and 6 of the present study showed that the dural branch of the AICA can be the main blood supply to SPS DAVFs.

The posterior meningeal artery originates from the PICA, OA, extradural vertebral artery, or ascending pharyngeal artery, and multiple anastomoses between these arteries are present. The artery of the falx cerebelli also arises from the PICA.

Multivariate analysis showed that location of the DAVF within the tentorium was an independent predictor for the existence of a pial arterial supply. This can be explained by the described rich supplies from dural branches of the PCA, SCA, AICA, and PICA to the tentorium including: the olfactory branches and the pericallosal branches originating from the ACA; the ADS from the PCA; the medial dural tentorial branch from the SCA; the subarcuate artery from the AICA; and the posterior meningeal artery and the artery of the falx cerebelli from the PICA.

Hypotheses Regarding Developmental Mechanisms of a "Pure" Pial Supply

In the present study, pial arterial supplies were classified as "pure" pial supplies in 8 cases. The pure pial supply cannot be explained by dilated pre-existing branches, and its angioarchitecture is different (corkscrew-like vessels, no evidence for a straight dural course, no known anatomical/cadaver correlates). One hypothetical mechanism that may be proposed for these vessels is induced angiogenesis, although no sequential

angiographic studies were available to verify this hypothesis in the present study. Several cases of newly developed DAVFs associated with a pial arterial supply after venous sinus thrombosis have been reported.[6,7] Angiogenesis induced by sinus thrombosis was proposed as a potential mechanism for the development of such pial arterial supplies.

A previously proposed mechanism of angiogenesis-induced shunts in DAVFs is through abnormal activation of vascular endothelial growth factor (VEGF)-mediated angiogenesis. In DAVFs, hypoxia due to the steal effect of a high-flow shunt and/or venous hypertension induces hypoxia-inducible factor-1 (HIF-1), which stimulates VEGF expression.[20–24] Several studies have reported that angiogenic factors including VEGF and HIF-1 were strongly expressed in both the dura mater sampled from patients with DAVFs and the cortex, pial vessels, and dura mater obtained from venous hypertension animal models.[21,23,24] In addition, induction of a pial arterial supply by hypoxia in patients with a vein of Galen malformation has been reported.[25] In support of this "venous hypertension–HIF–VEGF–angiogenesis" hypothesis is that, in the present study, the presence of venous dilatation (as a sign of venous hypertension) was a significant independent predictor of a pial arterial supply.

Younger age was another predictor of a pial arterial supply in the present study, and the mean age of patients with a pure pial supply was younger than that with dilated dural branches of a pial artery. While again only a hypothetical connection, studies on human muscle and in a murine leg ischemia model showed that capillarization and VEGF expression in response to stress were lower in older than in younger subjects.[26,27] In studies of moyamoya disease, development of indirect revascularization (pial synangioses) to ischemic brain was reported to depend on patient age.[28] While a weak link, this age-dependent angiogenetic response to hypoxia may explain younger age as a predictor for developing a pial arterial supply in the present study.

Treatment of DAVFs with Pial Arterial Supplies

No patients with a DAVF with a pial arterial supply in this series showed intracranial hemorrhage during or after treatment. A recent study reported that intracranial hemorrhage occurred during the embolization of DAVFs in 2 of 6 patients (33%) with an additional pial arterial supply to tentorial DAVFs.[4] In that study, the hemorrhagic complication rate was significantly higher in patients with a pial arterial supply than in those without, suggesting that the presence of a pial arterial supply is a risk factor for procedure-related hemorrhagic complications. Proposed mechanisms of immediate postinterventional hemorrhagic complications in patients with pial arterial supplies include associated (potentially occult) pial vascular malformation, feeder aneurysms, or fragility of neovascularized vessels induced by angiogenesis that will hemorrhage once the dural outflow is obliterated.[4,5,29,30] In the present study, postoperative bleeding did not occur even when the pial arterial supply was not obliterated, while the venous outflow was occluded. However, previous reports suggest a risk of hemorrhagic complications in the embolization of DAVFs with pure pial arterial supplies. Some studies suggested that obliteration of the pial arterial supplies before complete obliteration of the DAVF may lower the risk of intracranial hemorrhage. However, a pure pial arterial supply usually consists of tortuous and fragile vessels, and vessel rupture could occur during the catheterization and/or injection of embolic material. The retrospective nature of the present study, the small sample size, and the presumed relatively low risk of hemorrhage prevents us from making definitive recommendations regarding preoperative embolization of these vessels, despite the lack of hemorrhagic complications in the present series.

In the present series, no cerebral infarction caused by retrograde embolization through a pial arterial supply occurred. Preoperative understanding of the pial arterial supply to DAVFs is necessary to prevent comorbid cerebral infarction during procedures, especially when using a liquid embolic agent that can penetrate far into the involved vascular network. Careful procedures and knowledge of the anatomy are mandatory to avoid retrograde embolization into distal brain supplying pial arteries via the induced pial arterial supply during the embolization of DAVFs. Prior to the interventional treatment of a DAVF, careful scrutiny of the pial arterial supply to the DAVF is therefore essential.

Limitations of the Study

The most important limitation of the present study is its retrospective nature, with its associated absence of data on potential confounding factors, the (albeit small but potentially differential) loss to follow-up, and the lack of causal relationships. However, this study did have a sufficiently large comparison group to determine (a) the approximate prevalence of a pial arterial supply; (b) the type of pial supply present; and (c) the locations of DAVFs more likely to be associated with pial supplies. Still, since superselective angiography was not performed in all patients in this retrospective study, low flow or small additional pial arterial supplies may have remained undetected. Therefore, the presence of a pial arterial supply might have been underestimated in this series. No complications occurred in the treatment of DAVFs with a pial arterial supply in this series, but, as mentioned earlier, the small number, the inhomogeneous treatment strategies, and the presumed relatively low risk of hemorrhage contribute to a potential underestimation of hemorrhagic complications if a "pure" pial supply is not obliterated prior to the obliteration of the DAVF. A future prospective study of DAVFs with pial arterial supplies may show the mechanisms of complications caused by the pial arterial supply. Finally, this study was retrospectively conducted in patients over an 11-yr period, during which diagnostic and therapeutic devices and procedures developed; therefore, treatment bias could not be avoided.

CONCLUSION

The present retrospective study demonstrated a pial arterial supply in 23 of 204 patients with DAVFs (11.3%). Multivariate analysis showed that independent predictors of a pial arterial supply were younger age, DAVF located in the tentorium, and presence of venous dilatation. The results suggest that a pial arterial supply is related to the anatomic location of the DAVF, and it may suggest a potential angiogenetic association. Prior to the interventional treatment of a DAVF, recognition of a pial arterial supply to the DAVF is important to develop the treatment strategy, especially with respect to avoiding reflux into arteries supplying the brain.

Disclosure

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

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COMMENT

The authors describe a 10-year experience with 204 patients with dural arteriovenous fistulae (dAVF) and highlight the presence of "pial arterial supply" in 21 (10.3%) - which they categorically define as either previously described dural branches from pial vessels (ie, artery of Davidoff and Schechter) or "purely pial", presumably due to an aberrant angiogenic process akin to the characteristics often times observed in progressive, acquired dAVF associated with vein of Galen Malformations. They argue that "purely pial" lesions in particular predispose patients to peri-operative intracranial hemorrhage and that, accordingly, preoperative embolization is essential. Five examples of pial-supplied dAVF are provided, and statistical comparison to the rest of the patient cohort suggest slightly younger age, tentorium location, and venous dilatation

to be risk factors for pial arterial supply. All but 2 patients underwent some form of treatment, with 15 of them (71%) undergoing endovascular intervention, and an overall obliteration rate of 73.3% without any complications.

This is one of the first and only cohorts of dAVF patients with specific attention towards pial arterial supply, and the authors should be commended on addressing this important topic. The work is original, and sophisticated statistical analysis is performed (albeit on a small cohort of 21). The report alludes to a classification system for pial supply, but ultimately larger studies will be needed in order to validate the notion that "purely pial" lesions are indeed more problematic than dural branch-based pial contribution. This is difficult to do with rare diseases like dAVF, highlighting the need for multi-institution collaborations and bigger data sets. In addition, it is important to maintain diligent follow-up on these cohorts to ensure a durable treatment effect and report any instances of fistulae recanalization. Only then can investigators successfully recognize the complete impact that malignant features like pial arterial supply may have on the natural and treated histories of cerebrovascular pathology.

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