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Review Article

A Comprehensive Analysis of Dural Arteriovenous Fistula Involving the Superior Sagittal Sinus: A Systematic Review

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Objective: To evaluate published papers concerning dural arteriovenous fistula involving the superior sagittal sinus (SSS dAVF) treated with endovascular intervention.

Methods: A literature review was performed to identify studies reporting the clinical characteristics, vascular anatomy, endovascular techniques, embolic materials, angiographical result, and outcomes of SSS dAVF.

Results: The analysis consisted of 40 case reports or series, comprising a total of 51 cases of SSS dAVF treated with endovascular intervention. Clinical symptoms included hemorrhage (29.4%), infarction (17.6%), and seizure (7.8%). The arterial supply included the middle meningeal artery (MMA) (100%), superficial temporal artery (STA) (62.7%), and occipital artery (OA) (49.0%). Bilateral MMAs supplied in 78.4% of the cases. A pial arterial supply was observed in 21.6% of the cases. Stenosis or occlusion of the sinus was seen in 37.3% of the cases. The distribution of the Borden classification was as follows: I (7.8%), II (37.3%), and III (54.9%). The endovascular techniques were transvenous embolization (TVE) alone (11.8%), transarterial embolization (TAE) alone (74.5%), and a combination thereof (13.7%). The types of embolic materials for TAE were a coil (25.5%), n-butyl-2-cyanoacrylate (NBCA) (33.3%), and Onyx (45.1%). The arterial pedicles for TAE included MMA (86.3%), STA (17.6%), and OA (9.8%). Direct surgical exposure for the alternative endovascular access was performed to the SSS (5.9%), MMA (3.9%), STA (5.9%), and OA (3.9%). Sinus angioplasty with or without stenting was done in 9.8% of the cases. Balloon-assisted Onyx TAE was done with proximal flow control (7.8%), collateral devascularization (5.9%), and sinus protection (3.9%). Complete occlusion was achieved in 86.3% of the cases while the number of endovascular sessions varied among patients: single (74.5%), double (15.7%), and triple (9.8%). The rate of a postoperative modified Rankin Scale (mRS) score of 0–2 was 89.3%, with morbidity and mortality rates of 7.8% and 3.9%, respectively. Two patients died, possibly due to postoperative acute SSS occlusion.

Conclusion: The current systematic review disclosed several specific results, namely, the angioarchitectures of the SSS dAVF, the relationship between classification and hemorrhagic presentation, the diversity of treatment techniques, the association between the complete occlusion rate and the SSS condition, and the difficulty of achieving curable occlusion in a single session. These findings underscore the need for the development of endovascular techniques and devices to treat this challenging lesion. Improvements in adjunctive endovascular procedures, such as balloon-assisted techniques for Onyx TAE, may help support to the safe and effective obliteration of SSS dAVF.

Keywords ▶ dural arteriovenous fistula, endovascular intervention, superior sagittal sinus

Introduction

Dural arteriovenous fistula (dAVF) is an abnormal vessel communication between the meningeal artery and vein at the dura mater, often with recruitment of transosseous branches from the scalp arteries and less frequently associated with the pial branches from cerebral arteries. dAVF is the most commonly acquired lesion and is diagnosed in 0.29

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per 100000 adults per year,[1] and it rarely but occasionally involves the superior sagittal sinus (SSS), with a reported incidence ranging from 3.2% to 4.7% among cranial dAVFs.[1,2] A nationwide survey found a significant association between SSS dAVF and aggressive symptoms, including hemorrhage (OR 4.3, 95% CI: 1.6–9.6), venous infarction (OR 4.3, 95% CI: 1.7–10.0), and convulsion (OR 4.0, 95% CI: 1.5–9.8).[2] Aggressive intervention should therefore be considered to prevent or improve the symptoms of the disease, especially when cortical venous reflux exists.

Treatment of SSS dAVF generally involves endovascular intervention, open surgery, radiosurgery, or the combination thereof. In the last few decades, endovascular intervention in particular has been increasingly reported,[3–42] although most publications have been case reports or series, so the overview of this rare disease is still obscure. Accordingly, a literature review was conducted to identify studies reporting clinical characteristics, vascular anatomy, endovascular techniques, embolic materials, angiographic results, and clinical outcomes for the comprehensive analysis of the endovascular intervention of SSS dAVF. While several terms for this clinical entity were used in previous reports, such as dAVF involving the SSS and dAVF on the convexity adjacent to SSS, in this paper, we unified the term as "SSS dAVF" when applicable.

Methods

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) methodology guidelines were followed for this analysis.[43] A literature search was carried out by the author (SK) using PubMed and/or Google with the terms "dAVF" and/or "SSS" in English and Japanese. Relevant references from the selected articles were also included. Inclusion criteria for the analysis were as follows: (1) one or more patients with SSS dAVF undergoing endovascular intervention and (2) written in English or Japanese. Exclusion criteria for the analysis were as follows: (1) clinical studies in which the extraction of anatomical data of each patient was difficult and (2) lacking detailed description of endovascular intervention. The papers were screened by title and abstract to determine whether or not they met the inclusion criteria listed above. The two reviewers discussed any papers if there were controversial, and the third reviewers assessed these papers in a blinded manner. Any disagreements about inclusion and exclusion of the paper were resolved by consensus or mediation by the independent reviewer.

From each case report or series, detailed information, including the clinical characteristics, vascular anatomy, endovascular techniques, materials used, angiographic results, and clinical outcomes, were reviewed. Based on the drainage pattern, the lesions were classified according to both Borden and Cognard classification.[44,45] The arterial supply was categorized as follows: anterior falcine artery (AFA), ascending pharyngeal artery (APhA), meningohypophysial trunk (MHT), middle meningeal artery (MMA), occipital artery (OA), pial branch, posterior auricular artery (PAA), posterior meningeal artery (PMA), and superficial temporal artery (STA). The embolization method was categorized as transvenous embolization (TVE) or transarterial embolization (TAE). The embolic materials used were categorized as follows: particle, coil, n-butyl-2-cyanoacrylate (NBCA), and Onyx (eV3; Neurovascular, Irvine, CA, USA). The following outcome data were collected: angiographic results, periprocedural complications, and clinical outcome.

Categorical data were expressed as the proportion. Statistical analyses were considered inappropriate in this study due to the inconsistent measurement of the outcomes.

Results

The literature search obtained a total of 46 case series or reports, 6 of which were excluded following a review of the manuscript according to the exclusion criteria. Of the remaining 40 reports, a total of 51 patients met the inclusion criteria and were included in our analysis. The mean age (± standard deviation) of the patients was 55.1 ± 17.7 years, and 23.5% were females.

Table 1 summarizes the baseline characteristics and classification. Representative clinical symptoms included hemorrhage (29.4%), infarction (17.6%), and seizure (5.9%). The representative arterial supply was the MMA (100%), STA (62.7%), and OA (49.0%). Bilateral MMAs supplied in 40/51 (78.4%). A pial arterial supply was observed in 11/51 (21.6%). Stenosis or occlusion of the sinus was seen in 19/51 cases (37.3%). The distribution of the Borden classification was as follows: I (7.8%), II (37.3%), and III (54.9%). The distribution of the Cognard classification was as follows: I (5.9%), IIa (2.0%), IIb (13.7%), IIa+b (23.5%), III (23.5%), and IV (31.4%).

Table 2 summarizes the treatment and outcomes. The endovascular technique was subdivided into TVE alone (11.8%), TAE alone (74.5%), and TVE + TAE (13.7%). The types of embolic materials for TAE were coil (25.5%), NBCA (33.3%), and Onyx (45.1%). The representative

Table 1 Clinical and anatomical characteristics in the study cohort

Clinical and anatomical characteristics	Number (%)
Symptoms	
Hemorrhage	15/51 (29.4)
Infarction	9/51 (17.6)
Seizure	4/51 (7.8)
Other	24/51 (47.1)
Arterial pedicles	
MMA	51/51 (100)
STA	32/51 (62.7)
OA	25/51 (49.0)
AFA	9/51 (17.6)
PMA	9/51 (17.6)
MHT	5/51 (9.8)
AphA	3/51 (5.9)
PAA	1/51 (2.0)
Pial arterial supply	11/51 (21.6)
Stenosis/occlusion of SSS	19/51 (37.3)
Borden classification	
I	4/51 (7.8)
II	19/51 (37.3)
III	28/51 (54.9)
Cognard classification	
I	3/51 (5.9)
IIa	1/51 (2.0)
IIb	7/51 (13.7)
IIa + b	12/51 (23.5)
III	12/51 (23.5)
IV	16/51 (31.4)

AFA: anterior falcine artery; AphA: ascending pharyngeal artery; MHT: meningohypophysial trunk; MMA: middle meningeal artery; OA: occipital artery; PAA: posterior auricular artery; PMA: posterior meningeal artery; SSS: superior sagittal sinus; STA: superficial temporal artery

Table 2 Endovascular techniques and outcomes in the study cohort

Endovascular techniques and outcomes	Number (%)
Endovascular technique	
TVE alone	6/51 (11.8)
TAE alone	38/51 (74.5)
TVE + TAE*	7/51 (13.7)
Embolic material for TAE	
Coil	13/51 (25.5)
NBCA	17/51 (33.3)
Onyx	23/51 (45.1)
Arterial pedicles for TAE	
MMA	44/51 (86.3)
STA	9/51 (17.6)
OA	5/51 (9.8)
Direct surgical exposure	
SSS	3/51 (5.9)
MMA	2/51 (3.9)
STA	3/51 (5.9)
OA	2/51 (3.9)
Sinus angioplasty (± stenting)	5/51 (9.8)
Type of balloon assist for Onyx TAE	
Proximal flow control	4/51 (7.8)
Collateral devasculaization	3/51 (5.9)
Sinus protection	2/51 (3.9)
Angiographical outcome	
CO	44/50** (86.3)
Number of endovascular session	
Single	38/51 (74.5)
Double	8/51 (15.7)
Triple	5/51 (9.8)
Morbidity rate	4/51 (7.8)
Mortality rate	2/51 (3.9)
Postoperative mRS	
0–2	25/28 (89.3)
3–5	1/28 (3.5)
6	2/28 (7.1)
NA***	23/51 (45.1)

*Two cases of transarterial coil packing of the SSS were included. **Angiographical outcome was unavailable in one case due to a lack of detailed description. ***Clinical outcome was unavailable in 23 cases due to a lack of detailed description. CO: complete occlusion; MMA: middle meningeal artery; NA: not applicable; OA: occipital artery; SSS: superior sagittal sinus; STA: superficial temporal artery; TAE: transarterial embolization; TVE: transvenous embolization

arterial pedicles for embolization were MMA (86.3%), STA (17.6%), and OA (9.8%). Direct surgical exposure for the alternative endovascular access was performed to the SSS (5.9%), MMA (3.9%), STA (5.9%), and OA (3.9%). Sinus angioplasty with or without stenting was done in 5/51 (9.8%). Balloon-assisted Onyx embolization was done with proximal flow control (7.8%), collateral devascularization (5.9%), and sinus protection (3.9%).

Complete occlusion was achieved in 44/50 cases (86.3%) while the number of endovascular sessions varied among patients: single (74.5%), double (15.7%), and triple (9.8%). The overall rate of a postoperative modified Rankin Scale (mRS) score of 0–2 was 89.3%, with morbidity and mortality rates of 7.8% and 3.9%, respectively. Perioperative complications included facial palsy,[6] acute sinus thrombosis,[35] intracranial hematoma,[6,14] and scalp necrosis.[39] Two patients died possibly due to postoperative SSS occlusion.[6,35]

Table 3 summarizes the relationship between the classification and hemorrhagic presentation. The number of hemorrhagic presentation among Borden classification was as follows: I (0%), II (21.1%), and III (39.3%). The number of hemorrhagic presentation among Cognard classification was as follows: I (0%), IIa (0%), IIb, (28.6%), IIa + b (16.7%), III (0%), and IV (68.8%).

Table 4 summarizes the relationship between the outcomes and SSS condition. The rate of complete occlusion substantially

Table 3 Relationship between classification and the hemorrhagic presentation

Borden classification	No. of hemorrhage	Cognard classification	No. of hemorrhage
I	0/4 (0)	I	0/3 (0)
		IIa	0/1 (0)
II	4/19 (21.1)	IIb	2/7 (28.6)
		IIa + b	2/12 (16.7)
III	11/28 (39.3)	III	0/12 (0)
		IV	11/16 (68.8)

Table 4 Relationship between outcomes and SSS condition

	Stenosis/occlusion of the SSS	
	Yes (n = 19)	No (n = 32)
Endovascular technique		
TVE alone	3/19 (15.8)	3/32 (9.3)
TAE alone	10/19 (52.6)	28/32 (87.5)
TVE + TAE	6/19 (31.6)	1/32 (3.1)
Angiographical results		
Complete occlusion	13/19 (68.4)	31/32 (96.8)
Clinical results		
Postoperative mRS 0–2	NA*	25/28 (89.3)*

*Clinical outcome was unavailable in 23 cases due to a lack of detailed description. NA: not applicable; SSS: superior sagittal sinus

differed between SSS dAVF with or without stenosis/occlusion of the sinus (68.4% vs 96.8%, respectively).

Discussion

We evaluated the pooled data of 51 patients from 40 case reports or series for this systematic review to present the clinical manifestations and demonstrate the efficacy and safety of endovascular treatment for SSS dAVF. The current analysis disclosed several specific results: the angioarchitectures of the SSS dAVF, the relationship between classification and hemorrhagic presentation, the diversity of treatment techniques, the association between the complete occlusion rate and the SSS condition, and the difficulty of achieving curable occlusion in a single session. These findings underscore the need for the development of endovascular techniques and devices to treat this challenging lesion.

The patient demographics of the study cohort showed sexual and age-related tendencies, with an approximately threefold male dominance and a mean age of 55 years old, respectively. Similar tendencies were reported in a nationwide survey.[2)]

The representative presentations of the study cohort included hemorrhage (29.4%), venous infarction (17.6%), and convulsion (7.8%). The results almost agreed with a previous report that revealed symptoms of hemorrhage in 24%, venous infarction in 20%, and convulsion in 14%.[2)]

The distribution of hemorrhagic presentation in this study cohort was clearly stratified based on the Borden classification while it is important to avoid confusion with the natural hemorrhage rate. Furthermore, 11 (68.8%) out of 16 Cognard classification IV patients had a hemorrhagic presentation. Decompensation of an increased pressure load in a vulnerable venous structure (e.g., cortical vein) is presumably attributable to a hemorrhagic presentation in SSS dAVF.

We found that the dural and scalp arterial supply of the fistula was from the MMA in all cases and both the STA and OA in half of the cases, with other supply sources sporadically seen. A similar trend was reported in a previous study including 32 SSS dAVF: 97% from the MMA, 44% from the OA, 9% from the tentorial artery, and 3% from the APhA.[46)] Given its midline location, SSS dAVF frequently offers multiple branches from both external carotid arteries (ECAs).

Of note, one-quarter of SSS dAVFs had a pial artery supply, which is comparable to the rate found in a previous report.[47)] One study on 122 dAVFs at various locations found that, after treatment (including endovascular embolization in 70%) of the shunt, the occurrence of both posttreatment neurological deficits and cerebral infarcts differed significantly between the dAVFs with and without a pial artery supply (13.8% vs. 2.2% and 10.3% vs. 1.1%, respectively).[47)] Treatment decisions and therapeutic strategies for SSS dAVF with a pial arterial supply remain problematic. Gamma knife radiosurgery may be an alternative option for managing such challenging lesions.[48)]

TVE of the affected sinus was performed in one-quarter of SSS dAVF cases. Traditionally, TVE has been thought to safely occlude the entrance of drainage in cases with sinus drainage (i.e., lacking a direct cortical vein reflux). Selective[12,41,49,50)] and transarterial[13,21)] coil packing have also been reported. On the other hand, stenosis or occlusion of the affected sinus was identified in one-fifth of the cases. In such situations, the utility of surgical exposure of the SSS or direct puncture for alternative access to the affected sinus has been reported.[6,31,35,51)]

Reopening of the occluded sinus secondary to SSS dAVF has also been reported.[23,29,38,49,52] A transvenous reopening technique with gentle rotational advancement of a 0.035-inch guidewire for subsequent microcatheter navigation was successful for managing 53 (82%) out of 62 cases of occluded sinus.[50] However, among 51 of the patients with a reopened sinus in this report, 5 (9.8%) presented with complications, such as vessel perforation and subdural hematoma.

Few reports have described balloon sinus angioplasty with or without stent placement.[29,35,42] Stent placement may prevent the early collapse of the recanalized sinus and secure durable patency, although its clinical validity has not been verified. Navigation of the unyielding system through the transverse-sigmoid junction may not be technically feasible.

In the past two decades, the role of TAE for SSS dAVF has changed steadily, largely owing to improvements in embolic materials. Particles and coils are typically used to achieve provisional flow reduction, which is a palliative strategy and is not intended to obtain permanent occlusion. Additionally, it has been indicated that proximal occlusion with coils may miss the chance to achieve complete occlusion by losing an accessible arterial pedicle.[23] On the other hand, liquid embolic agents, such as NBCA and Onyx, have the potential to achieve curable occlusion by ensuring the effective progression of the agent toward the fistula. One study compared the angiographic results of SSS dAVF before and after the introduction of Onyx. Initial angiographic occlusion was achieved in 6 out of 10 cases (60%) before the Onyx era, whereas it was achieved in 18 out of 22 cases (82%) in the Onyx era.[46]

MMA was selected as the arterial pedicle for TAE in the vast majority of the cases, although its anatomical course and vessel caliber varied among patients. In cases where the best or only arterial pedicle for TAE is endovascularly inaccessible due to the tortuosity of the proximal part of the artery, surgical exposure, and direct puncture of the distal part of the vessel has been attempted using the MMA,[10,28] STA,[6,10,28] and OA.[6]

With regard to Onyx TAE, balloon-assisted techniques are anticipated to expand the application range of this liquid, non-adhesive agent. In general, these approaches include balloon devascularization of the collateral arterial branches,[22,23,40] proximal flow control of the injecting artery,[20,37,40] and protection of the functioning but compromised sinus.[14,35] The details of each technique have been described previously.[23]

The current analysis revealed a complete occlusion rate of 86.3%, postoperative mRS score of 0–2 in 89.3%, and morbid and mortality rates of 9.1% and 7.8%, respectively. A previous study described the outcomes of 32 SSS dAVFs following endovascular intervention, noting an initial angiographic occlusion rate of 75%, overall complication rate of 16%, and permanent neurological deficit rate of 0%.[46] While the direct comparison of the outcomes is inappropriate due to the differences in the clinical backgrounds between studies, these results of endovascular intervention are encouraging for future studies.

Our results suggested that the complete occlusion rate might be influenced by the SSS condition. Further study is needed to clarify this point.

Our analysis highlighted several issues. First, the possible cause of death in the two dead cases was acute occlusion of the SSS postoperatively.[6,35] Clarifying the occlusive mechanism and securing the functional venous circulation may be required. Second, achieving complete occlusion of SSS dAVF in a single session was still challenging in some cases. Further improvements in endovascular techniques and devices are therefore needed. Third, although SSS dAVF carries a higher hemorrhagic risk than dAVFs at other locations, whether or not endovascular treatment actually prevents hemorrhage in the long-term follow-up is unclear. Approximately half of all cases lacked a sufficient description of the clinical outcome. In addition, details regarding the venous angioarchitecture, such as sinus or non-sinus type, and the coexistence of a localized shunted venous pouch (parasinus), were also frequently insufficiently described. Finally, it must be mentioned that a systematic review cannot fully exclude a positive results bias, as negative results are less frequently published than positive ones. A prospective approach will help resolve such shortcomings.

Conclusion

The current systematic review disclosed several specific results: the angioarchitectures of the SSS dAVF, the relationship between classification and hemorrhagic presentation, the diversity of treatment techniques, the association between the complete occlusion rate and the SSS condition, and the difficulty of achieving curable occlusion in a single session. Our findings underscore the need for the development of endovascular techniques and devices to treat this challenging lesion. Improvements in adjunctive endovascular procedures, such as balloon-assisted techniques for Onyx TAE, may help support to the safe and effective obliteration of SSS dAVF.

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Disclosure statement

The authors declare no conflicts of interest associated with this manuscript.

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