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ORIGINAL ARTICLE - VASCULAR NEUROSURGERY - ARTERIOVENOUS MALFORMATION

Cranial dural arteriovenous shunts: selection of the ideal lesion for surgical occlusion according to the classification system

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Abstract

Background The types of cranial dural arteriovenous fistulae (cDAVFs) that constitute good surgical candidates are unclear despite the use of classifications. We aimed to compare the DES classification with other classification schemes in identifying "ideal lesions for surgery." The DES scheme is based on two features: the level of the shunt (BVS, bridging vein shunt; DSS, dural sinus shunt; ISS, isolated sinus shunt; EVS, emissary vein shunt) and the type of leptomeningeal venous reflux (LVR) (direct, exclusive, strained).

Methods In this observational cohort study, the angiographies of 20 consecutive patients treated over 1 year were analyzed retrospectively. We defined cDAVFs as ideal for surgery, if cure may be achieved by disconnecting the arterialized draining vein through a single craniotomy. To evaluate the performance of each classification scheme in identifying the "ideal lesion for surgery," we carried out a sensitivity analysis of the Borden, Cognard, and DES schemes.

Results Eight lesions were Borden type 3 and 1 type 2, and 11 type 1. According to Cognard, 2 lesions were type IV, 2 type III, 1 type IIa+b, 11 type I, and 4 lesions could not be clearly classified. According to the DES scheme, 8 lesions were DSS, 4 BVS, 3 ISS, and 5 EVS. All 4 lesions classified as BVS in the DES were ideal lesions for surgery (sensitivity, specificity, PPV, NPV 100%). Not all high-grade lesions according to Borden were good surgical candidates.

Conclusion The DES scheme, as opposed to other classifications, facilitates the therapeutic decision-making especially for selecting candidates for surgery.

Keywords Arteriovenous · Classification · Cranial · Dural · Fistula · Shunt

Introduction

As a rule, cranial dural arteriovenous fistulae (cDAVFs) are acquired lesions affecting one of the following venous structures: the dural sinus wall (dural sinus shunt), the transdural segment of the bridging veins (bridging vein shunt), or the emissary and epidural veins (emissary vein shunt) [4, 5]. Whereas leptomeningeal venous reflux is an inherent characteristic of all bridging vein shunts, the dural sinus shunts frequently and the emissary vein shunt infrequently present leptomeningeal venous reflux, which is due to their specific venous angioarchitecture.

Non-specific terms such as "high-grade" or "high-risk" cDAVFs have often been used in recent years to describe lesions with leptomeningeal venous reflux. The surgical disconnection of the vein carrying the reflux is a highly effective and straightforward strategy to treat cDAVFs presenting reflux and is applied for many years [12, 15, 18, 27]. However, careful reading of the published reports shows that the definition of a high-grade cDAVF remains subjective and is not consistent across the main body of the literature. Some authors [2, 21, 22] account that "high-grade" simply defines lesions with any leptomeningeal venous reflux, whereas other authors

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[2, 17, 18, 21, 22] include only the cDAVFs Borden [8] type 3 or Cognard [10] type III and IV and not the Borden type 2 or Cognard type IIb and type IIa+b.

These discrepancies and confusion are further enhanced by the fact that the definition of Borden type 3 lesions includes not only the direct shunts into a bridging vein (bridging vein shunt) but also the isolated sinus shunts, which are located in the wall of an isolated dural sinus [8]. The isolated sinus shunts, however, are not clearly classified as Cognard type III lesions [10], which in general appear as equivalent to the Borden type 3 lesions in most publications [9, 14, 16, 28, 31].

Recently, a new classification system of cDAVFs, the DES scheme [6], has been proposed, based on two features: first, the precise anatomical localization of the level of the shunt (bridging vein shunt, dural sinus shunt, isolated sinus shunt, and emissary vein shunt) and, second, the characteristics of the leptomeningeal venous reflux (direct, exclusive, strained; DES) [6]. The purpose of the current study is to test the hypothesis that the precise anatomical localization of the shunt, as used in the DES scheme, facilitates the selection of cDAVFs, which are ideal for surgical occlusion and for which surgery represents an attractive alternative to embolization. To obtain this, we compared the DES scheme with the two most used classification systems, the Borden and the Cognard, with regard to the usefulness in therapeutic decision-making.

Methods

This is an observational cohort study, which included 20 consecutive patients with cDAVFs diagnosed and treated over a period of 12 months, from May 2014 to April 2015 at our hospital. Their angiographies were retrospectively reviewed, and Borden and Cognard grades as well as DES features concerning anatomical localization were assigned.

We defined "high-grade" cDAVF in 2 different ways: *definition 1* being any cDAVF with leptomeningeal venous reflux (therefore Borden grade 2 and 3 as well as Cognard grade IIb, IIa+b, III, and IV) and *definition 2* being only the cDAVF Borden grade 3, respectively Cognard grade III or IV. At our institution, the management protocol included embolization as the first therapeutic option. Embolization was therefore proposed and attempted for all patients. Surgery was performed only after failure of embolization or necessity for acute hematoma evacuation.

For this study, we decided to define the "ideal cDAVFs for surgical treatment," as a cDAVF that can be completely occluded by direct disconnection of the arterialized draining vein through a "single-approach" craniotomy. Craniotomies that expose both the supra- and infratentorial region or both left- and right-sided regions were not considered "single-approach" craniotomies. Therefore, lesions with multiple draining veins requiring extended approaches or skeletonization of the sinus were not considered ideal surgical cDAVFs. This definition is arbitrary and reflects our strategy towards minimally invasive surgery. Furthermore, it was defined in retrospect, which means that the concept of ideal surgical lesion did not necessarily imply that those patients were actually treated by surgery. According to our policy of first option endovascular treatment in use during the period of this review, ideal surgical candidates still may have been treated by endovascular embolization.

Detailed analysis of the venous angioarchitecture of each "high-grade" lesion was performed to determine which cDAVFs fulfilled the criteria of ideal surgical lesion (Y/N).

The quality of imaging was high as well as the completeness of information on venous anatomy. We opted to study a non-selected cohort of patients over a short period of 1 year, in order to illustrate, through real-life practice, the ability of classification systems to guide the management and surgical selection of patients with cDAVFs.

To evaluate the performance of each classification in identifying the cDAVFs judged to be ideal surgical lesions, a sensitivity analysis was carried out for both groups, according to definitions 1 and 2 of "high-grade" cDAVFs. However, Cognard classification was not compared further as the grading was not clear in 4 patients (two IIb or IIa+b or III, one IIa+b or III, and one IIb or IIa+b or IV). This is inherent to the Cognard grading system [6, 10, 24]. Therefore, we calculated sensitivity and specificity of the Borden and DES classifications (categorical variables) in identifying the ideal candidate for surgery using 2×2 tables, as well as their positive and negative predictive values.

Results

Ten lesions were located in the transverse and sigmoid sinus. Two lesions were located in the cavernous sinus, 2 in a medullary bridging vein, 1 in an inferior vermian bridging vein, 1 in a petrosal bridging vein, 1 in the jugular bulb, and 3 in the hypoglossal emissary vein. Twelve patients presented with tinnitus, 3 with hemorrhage, 1 with neurological deficit, 1 with epilepsy, 1 with congestive orbital symptoms, and 1 with headache, and 1 was incidental.

Correlation between "high-grade" cDAVF and ideal surgical lesion

Of the 20 patients treated during the study period, 9 (1–9) had leptomeningeal venous reflux and therefore were "high-grade" shunts according to definition 1 (Borden grade 2 and 3), whereas 8 patients (1–8) harbored "high-grade" cDAVFs according to definition 2 (Borden grade 3).

Among the 9 patients with "high-grade" cDAVFs according to definition 1, patients 2, 3, 4, and 6 (4 out of 9; 44%)

fulfilled the criteria of ideal surgical lesions (Y) (Fig. 1) and the rest of the patients did not (N) (Fig. 2). The same analysis on the 8 patients corresponding to definition 2 for "high-grade" cDAVFs showed the same patients 2, 3, 4, and 6 (4 out of 8; 50%) fulfilling the criteria for ideal surgical lesion (Table 1). All four cDAVFs (patients 2, 3, 4, 6) fulfilling the criteria of ideal surgical lesions were Borden type 3. However, 4 cDAVFs (patients 1, 5, 7, 8) who did not fulfill the criteria of ideal surgical lesions also were Borden type 3. Patient 9 had leptomeningeal venous reflux and harbored a Borden type 2 lesion, and did not fulfill the criteria for ideal surgical lesion either. Therefore, among the patients with "high-grade" cDAVFs, neither definition 1 nor definition 2 allowed to clearly separate cDAVFs ideal for surgical treatment. Figures 1 and 2 show two patients illustrating this fact.

Correlation between DES grade and ideal surgical lesion

The same analysis using the DES scheme showed that all the cDAVFs fulfilling the criteria for ideal surgical lesions were bridging vein shunts (patients 2, 3, 4, 6). Conversely, among the patients who did not fulfill the criteria for an ideal lesion for surgery, none had a bridging vein shunt, 4 had an isolated sinus shunt, and 1 had a dural sinus shunt (patients 1, 5, 7, 8, 9).

Outcomes of diagnostic analyses

The sensitivity of "high-grade" type for detecting the ideal lesions for surgery was 100% and its specificity 0%, for both definitions. Its positive predictive value was 44% for definition 1 and 50% for definition 2, and its negative predictive value 0% for both. In comparison, the sensitivity and specificity as well as positive and negative predictive values of the DES type for identifying ideal surgical cDAVFs all were 100% (Supplemental digital content 1. Appendix table 1).

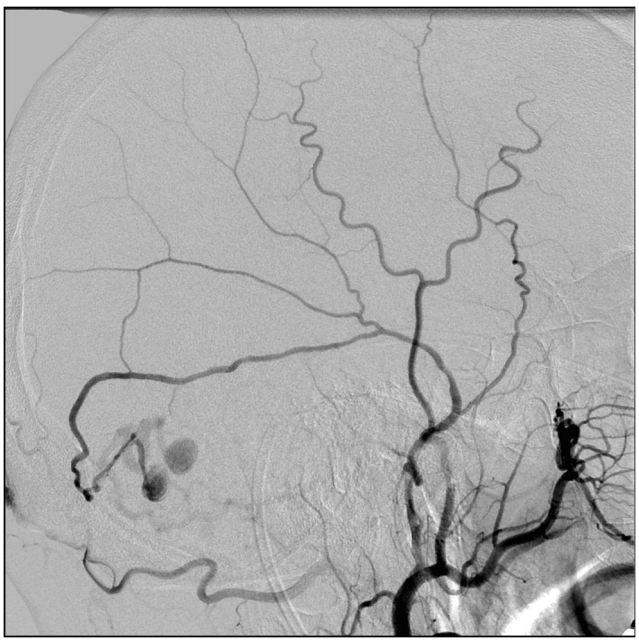

Fig. 1 Case No. 2. Typical example of a bridging vein shunt of a tentorial bridging vein with DES (direct, exclusive, and strained) leptomeningeal venous reflux. Borden grade 3 and Cognard grade IV

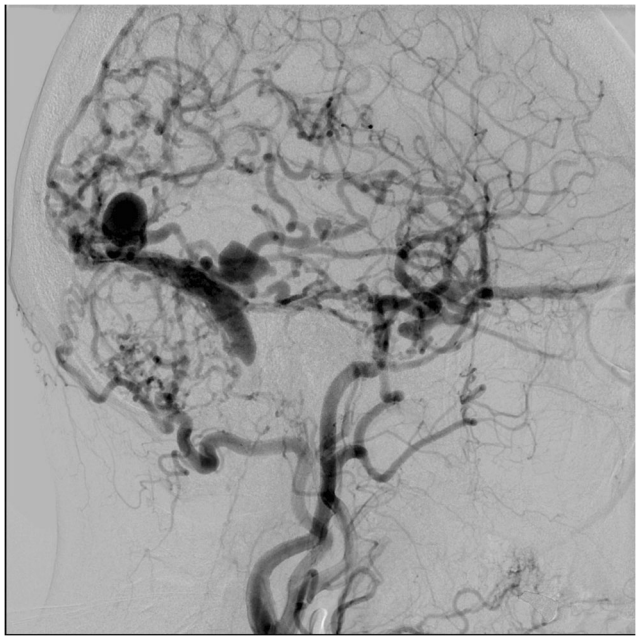

Fig. 2 Case No. 8. Typical example of an isolated sinus shunt with non-direct (nD) but exclusive (E) leptomeningeal venous reflux with significant, mostly ectatic, strain (S) of the cortical venous system. Borden grade 3. Also, a typical example of the ambiguity of the Cognard classification. It is unclear if the lesion is grade IIb due to anterograde (?) flow into the isolated sinus segment, IIa+b due to retrograde (?) flow into the isolated sinus segment, or IV due to ectasia of the cortical draining veins

Treatment modalities

Since our management strategy was to offer the endovascular approach as first option, 17 patients out of 20 were treated with embolization. Among the 4 cDAVFs fulfilling the criteria of ideal surgical lesions, embolization was performed in 2 (patients 3, 6), whereas the other two were treated by surgery: patient 2 was operated for an acute hematoma evacuation and patient 4 after he failed endovascular attempt (Table 1). Both these patients had a cDAVF Borden grade 3, and both were bridging vein shunts according to the DES.

Patient 8 was treated by a combined surgical and endovascular approach. The patient underwent surgery to evacuate a hematoma; however, the cDAVF was not an ideal lesion for surgical interruption. He carried a lesion Borden grade 3, and the DES scheme indicated an isolated sinus shunt and not a bridging vein shunt (see description below). Interestingly, a comparison between the cDAVFs treated by a combination of surgery and embolization (patient 8) with

Table 1 The patients with high-grade cDAVF according to definition 1 (1–9) and definition 2 (1–8). Location of the shunts. Ideal surgical candidates, Y yes, N no. Classification according to Borden, Cognard, and DES scheme. *BV* bridging vein, *LVR* leptomeningeal venous reflux, *BVS* bridging vein shunt, *DSS* dural sinus shunt, *ISS* isolated sinus shunt, *EVS* emissary vein shunt, *D* direct, *E* exclusive, *S* venous strain, *n* non

Pt	Location	Ideal surgical candidate	Borden	Cognard	DES	
					Level	LVR
1	Transverse	N	3	IIb, IIa+b, or III	ISS	nDES
2	Tentorial BV	Y	3	IV	BVS	DES
3	Tentorial BV	Y	3	III	BVS	DES
4	Tentorial BV	Y	3	III	BVS	DEnS
5	Transverse	N	3	IIb, IIa+b, or III	ISS	nDEnS
6	Medullary BV	Y	3	IV	BVS	DES
7	Transverse	N	3	IIa+b or III	ISS	nDEnS
8	Transverse	N	3	IIb, IIa+b, or IV	ISS	nDES
9	Transverse	N	2	IIa+b	DSS	nDnES

those treated by surgery alone (patients 2 and 4) showed significant differences in venous angioarchitecture. Patients 2 and 4 (both bridging vein shunts) were treated completely by a small single craniotomy and direct disconnection of the draining vein and therefore in retrospect fulfilled the criteria of ideal surgical lesions. To the contrary, patient 8 (isolated sinus shunt) was first treated for surgical hematoma evacuation and disconnection of two supratentorial arterialized draining veins, which constituted the source of bleeding. However, the cDAVF had additional infratentorial draining veins, which would have required a more extended surgical approach and therefore, an endovascular approach was decided instead to complete the treatment. This cDAVF (isolated sinus shunt) was not an ideal lesion for surgical interruption, even though it was a Borden type 3.

Discussion

This study includes the whole spectrum of cDAVF types and provides a vivid demonstration of the differences among the classification systems, especially in regard to the microsurgical treatment decisions. Our study showed a higher sensitivity and specificity as well as negative and positive predictive values for the DES scheme as compared with the Borden system in defining ideal surgical candidates. The Cognard classification could not be analyzed due to ambiguity in grading some cDAVFs. Similarly, the criteria used in the literature for the definition of "high-grade" cDAVFs were not helpful in predicting which cDAVFs are ideal lesions for microsurgical treatment.

Microsurgery can cure most of the cDAVFs [30]; however, in an era in which endovascular approaches appear as the treatment of choice for fistulae involving most [21, 22, 26] if not all [1, 3, 11] intracranial locations, the crucial question is not merely the feasibility of a treatment, but rather the selection of the treatment with the highest efficacy and the lowest morbidity and mortality [13, 25]. In other words, the important question is correct patient selection, in particular the reliable identification of those cDAVFs for which surgery offers a highly effective occlusion and constitutes an attractive treatment alternative to embolization.

In the contemporary neurosurgical literature, a clear definition of the cDAVFs that constitute a good surgical lesion is missing or appears nebulous. The first publications, which described the technique and demonstrated the efficacy of simple disconnection of the draining vein, were the ones by Grisoli et al. [15] who described the treatment of four tentorial cDAVFs and subsequently by Thompson et al. [27] who reported on the surgical interruption of a single draining vein in another four patients and later by Collice et al. [12] who made a step forward and described 20 patients with "pure leptomeningeal venous drainage." They differentiated them clearly from the isolated sinus shunt variant, even though they still considered the sinus wall the true location of these shunts and not the transdural segment of the bridging vein (bridging vein shunt), as recently described [4]. Interestingly, these authors did not use the Borden nor the Cognard system to describe the lesions. The likely explanation is that both these classification systems of cDAVFs have inherent limitations from the point of view of surgical treatment decision by not specifically reflecting the venous angioarchitecture. Indeed, Collice et al. [12] found both classifications unpractical and did not use any of the two for the characterization of the lesions they reported. This is also the reason why vague terms such as "high-grade" or "high-risk" cDAVFs with discrepant definitions have been used in the surgical literature to describe the performance, indication, or strategy of treatment during the last years. In fact, under the term "high-grade," some authors have included all lesions with leptomeningeal venous reflux, i.e., lesions

Borden type 2 and 3 [2, 20–22, 31], whereas others have meant only lesions Borden type 3 [17–19, 23].

If we consider all cDAVFs with leptomeningeal venous reflux (according to definition 1 of "high-grade") potential lesions for surgical treatment, we immediately realize that this criterion cannot select ideal surgical candidates, as Borden type 2 lesions fundamentally differ from type 3 lesions (compare Figs. 1 and 3). A cDAVF Borden type 2 drains into both the dural sinuses and the cortical veins (reflux). In type 2 lesions, the draining cortical veins are often multiple and their surgical identification and disconnection are usually tedious. Furthermore, we have to be aware that venous disconnection alone does not cure these lesions. Disconnection of the draining veins simply transforms a Borden grade 2 cDAVF into a grade 1 lesion [29]. Such a strategy can be an option in some patients but is not the ideal strategy. Therefore, we believe that it is fair to state that many of the "high-grade" shunts according to definition 1 are not primarily ideal surgical candidates.

If we consider all Borden type 3 cDAVFs (according to definition 2 of "high-grade") potential lesions for surgery, we also realize that the lesions within this group are heterogeneous (compare Figs. 1 and 2). This is related to the original definition of Borden type 3, which includes both the shunts of a cortical vein with direct leptomeningeal venous reflux as well as the shunts of an isolated sinus variant with non-direct leptomeningeal venous reflux [8]. What makes these group heterogeneous are the differences in localization of the shunt as well as their venous architecture. The lesions with direct shunt into the cortical vein do not involve the sinus and are located at the junction of the cortical vein with the sinus (bridging vein). As a rule, the draining vein is single and not connected to the lumen of the sinus. On the contrary, the isolated sinus shunt variant drains first into the sinus and only secondarily retrogrades into the cortical vein (non-direct). The shunt is located in the sinus wall, and as a rule, the draining veins are multiple and often involve both supratentorial and infratentorial veins (typically for the transverse sinus) or bilateral veins (typically for the superior sagittal sinus). The surgical strategy to achieve a complete disconnection is therefore more challenging and usually more tedious than the disconnection of a single vein of a bridging vein shunt with direct leptomeningeal venous reflux. Therefore, we believe that it is fair to state that some "high-grade" cDAVFs defined as Borden type 3 are not primarily ideal surgical candidates.

The DES scheme [6] is based on precise description of the anatomical localization of the level of the shunt (bridging vein shunt, dural sinus shunt, emissary vein shunt) and the type of leptomeningeal venous reflux (direct, exclusive, strained: DES). A dural sinus shunt that drains exclusively through the cortical veins (no venous outflow through the sinus itself) is classified as an isolated sinus shunt [6]. All types of shunts may present leptomeningeal venous reflux. Bridging vein shunts and isolated sinus shunts always have reflux by definition, whereas dural sinus shunts and emissary vein shunts may present with or without leptomeningeal venous reflux [6]. The reflux is further characterized by its directness and exclusivity as well as the induced venous strain. Among all types of cDAVFs with reflux, the presence of venous strain identifies the lesions with the most aggressive mode of clinical presentation (bridging vein shunt with strain Fig. 1 > isolated sinus shunt with strain Fig. 2 > dural sinus shunt with strain Fig. 3), and among the cDAVFs with reflux, but without venous strain, the bridging vein shunts have the more aggressive mode of presentation (bridging vein shunt without strain Fig. 4) [7].

The understanding of the precise shunt localization and the venous angioarchitecture is mandatory to determine the most adequate treatment strategy. Patients harboring a bridging vein shunt type of cDAVF carry an ideal lesion for surgical cure, as the fistula shunts directly into a single leptomeningeal vein at the junction with the dura, offering a single and precise spot for surgical interruption of the shunt. Comparing the different venous angioarchitectures, it becomes clear that patients

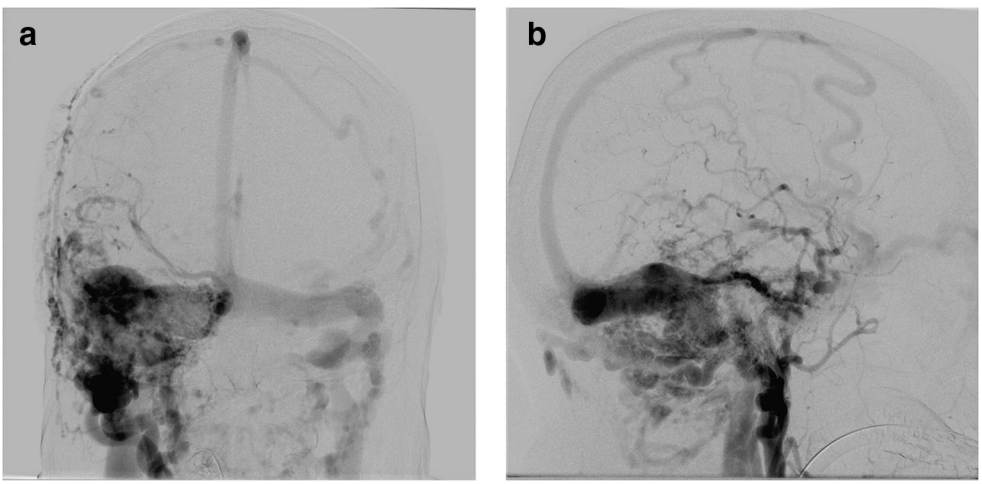

Fig. 3 Case No. 9, **a** anteroposterior and **b** lateral view. An example of a dural sinus shunt with nDnES reflux, Borden grade 2 and Cognard grade IIa+b

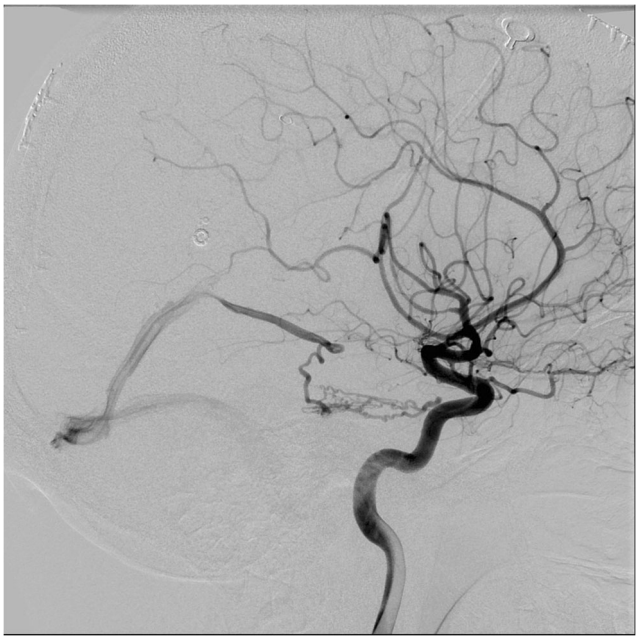

Fig. 4 Case No 4. Typical example of a bridging vein shunt with DEnS reflux. Cognard grade III. The Borden grade is also 3, identical with case No. 2 and case No. 8

harboring bridging vein shunts constitute ideal candidates for surgery. The purpose of any classification system is to reflect the disease in a reliable, reproducible, and clinically useful manner. The 100% sensitivity, specificity, and positive and negative predictive values for our scheme found in this small series tend to corroborate the value of the DES scheme in clinical decision-making.

Limitations of the study

An obvious limitation of the above statement is the appropriate reading of the angiographic investigations, which may be difficult in some complex cDAVFs and therefore may introduce information bias. The DES scheme obliges the physician to perform a very accurate analysis, in order to identify the precise localization of the level of the shunt and the features of the leptomeningeal venous reflux. As the DES classification is not known as broadly as the Borden, the reader may find it helpful to consult the schematic drawings presented in the appendix (Supplemental digital content 2. Figure A, B, C, D) and consult the original description of 2014 [6]. Correct interpretation of the angioanatomy of any vascular malformation remains the main step before any treatment planning.

A further limitation of our study is the arbitrary definition of "ideal surgical cDAVF" (see "Methods"). We believe that, whenever alternative treatment options are available, each method must critically analyze its selection criteria in order to minimize the invasiveness of a treatment and at the same time optimize the therapeutic efficacy. In the "Methods" section, we describe the reasons for the definition of "ideal lesion for surgical cure." We wanted a simple definition, in line with the tendency to favor minimally invasive surgical strategy. Finally, our management protocol for cDAVFs during the period under review defined endovascular embolization as the first treatment option. Therefore, two patients that had ideal surgical lesions underwent endovascular cure. Ideal for surgery does not imply that endovascular treatment is not feasible; similarly, microsurgery can also cure other cDAVFs. The purpose of this study was not to compare treatment outcome but to compare the classification systems for surgical decision-making of cDAVFs.

Conclusion

This study illustrates the usefulness of the DES scheme for treatment decision-making. Patients with cDAVFs diagnosed as bridging vein shunt type, regardless of the presence or absence of venous strain, constitute good candidates for microsurgical shunt interruption. A prerequisite is the correct interpretation and understanding of the angioarchitecture of the cDAVF, which can be challenging in some complex shunts.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (name of institute/committee) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. An Ethics committee approval was obtained (KEK 2016-02126), and all individual participants included in the study signed an informed consent.

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