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Cranial dural arteriovenous shunts. Part 3. Classification based on the leptomeningeal venous drainage

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Abstract The commonly used Borden and Cognard classification systems for the prediction of clinical behavior of cranial dural arteriovenous shunts focus on the venous drainage, particularly the presence of leptomeningeal venous drainage, and on the direction of flow, particularly the presence of retrograde flow. In addition, the latter includes ectasia and spinal drainage as criteria of two distinct grades. However, none of the above classifications (a) differentiates direct from exclusive leptomeningeal venous drainage, (b) considers cortical venous congestion as a factor potentially associated with an aggressive clinical course, and (c) anticipates ectasia in shunts with a mixed dural-cortical venous drainage (type 2). In this study, we analyzed the angiographic images of 107 consecutive patients having a cranial dural arteriovenous fistula with leptomeningeal venous drainage, based on a newly developed scheme. This scheme, symbolized with the acronym "DES," groups the dural shunts according to three factors: *directness* and *exclusivity* of leptomeningeal venous drainage and signs of venous *strain*. According to the combination of the three factors, eight different groups were distinguished. All analyzed cases could be assigned to one of these groups. Directness of leptomeningeal venous drainage expresses the exact site of the shunt (bridging vein vs sinus wall), whereas exclusivity expresses venous outlet restrictions. All bridging vein shunts had a direct leptomeningeal venous drainage. Almost all bridging vein shunts and all "isolated" sinus shunts had an exclusive leptomeningeal venous drainage. Venous strain, manifested as ectasia and/or congestion, denotes the decompensation of the cerebral venous system due to the shunt reflux. The comparison of the presented concept with the currently used classifications highlighted the advantages of the former and the weaknesses of the latter.

Keywords Dural arteriovenous shunts · Arteriovenous fistula · Leptomeningeal drainage · Cortical venous drainage · Retrograde flow · Classification

Introduction

Several different or similar classifications have been proposed over the years for the analysis of cranial dural arteriovenous fistulae (CDAVF) [1–8]. Two of these classification schemes dealing with the venous drainage in relation to the clinical presentation are mostly used and cited in the literature [1, 2]. All classifications in medicine are supposed on the one hand to be easy and practical, facilitating research and communication, and on the other to reflect the described aspects of the entity as precisely as possible. Then, it is the duty of the analyzing physician to apply correctly the criteria of each classification in order to achieve meaningful and reproducible conclusions. However, as failure to comprehend the essential features of the disease may masquerade as erroneous application of a classification, similarly a classification scheme can sometimes hinder the comprehension of these features [9]. No matter how much good a classification is, it is still a code, and as all codes, it is linked to simplifications and generalizations under the concrete features described in the scheme, providing

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symbols and substituting real individual characteristics. In other words, each classification inevitably has inherent limitations, which may become significant especially in cases of subgroup analysis.

In the literature, the terms "direct" and "exclusive" leptomeningeal venous drainage (LVD) are often used without discrimination to describe the concept of exclusivity [1, 10] or not used at all [8].

We aimed to analyze our cases of CDAVF with LVD focusing on three important characteristics: the directness and exclusivity of LVD and the strain of the leptomeningeal venous system. The formed groups were compared with the groups based on the Borden and Cognard classifications. In this process, the weaknesses of the latter classification systems became evident.

Materials and methods

From a series of 211 consecutive patients with CDAVF treated in our department during the last 19 years, the cases with complete angiographic images, which presented LVD, were retrospectively analyzed [11]. Cases of pediatric and/or congenital CDAVFs were excluded. Based on angiographic imaging, the exact location of the shunt and relationship to the draining leptomeningeal vein(s) were recorded. Since the LVD is considered the decisive angioarchitectural feature that determines the aggressive nature of the shunt, each lesion was grouped according to the criteria of directness and exclusivity of the LVD. A direct LVD was defined by venous drainage that used the bridging and leptomeningeal vein(s) without interposition of any sinus. In these cases, the exact location of the shunt was the bridging vein (BV) and not the venous sinus. A nondirect LVD was defined by venous drainage that used the BVs but with the interposition of a sinus, which implies that the shunt was primarily or solely located in the wall of the sinus. An exclusive LVD was defined as venous drainage by only the leptomeningeal veins either because the shunt was located in a BV with its exit to the sinus occluded or because the only exit of the sinus was through the BVs to the leptomeningeal venous system. A nonexclusive LVD was defined by drainage that occurred both by cortical veins and also by the venous sinuses, dural veins, or emissary veins (EV). Presence of ectasias or congestive pseudophlebitic appearance was recorded as cortical venous strain. Other associated venous outflow restrictions (VOR) of the venous sinuses, as complete or partial thrombosis or stenotic appearance, were also recorded. In cases of multiple shunts, only the higher-grade lesion was studied.

Results

One hundred and seven patients with LVD were identified. The possible combinations of the above criteria defined the groups of lesions presented in the Fig. 1. An imaginary venous sinus was used for the illustrative demonstration of the described dural shunt groups (Fig. 2).

All CDAVFs could be analyzed according to the DES concept (*directness* and *exclusivity* of LVD and venous *strain*. The letter "n" stands for non/no, e.g., nD-nE-nS: nondirect LVD, nonexclusive LVD, no venous strain). The nD-nE-nS group included 23 cases, all with lesions of Borden type 2 and Cognard type IIb or IIa + b (Fig. 3a, b). The nine cases of the nD-nE-S group were lesions that according to the criteria of venous drainage and direction of flow could be classified as Borden type 2 and Cognard type IIb or IIa + b (Fig. 4a, b). Additionally, all of these patients had signs of venous strain, features, which cannot be recorded by the above classifications. Two patients had a venous ectasia, six had a congestive pseudophlebitic venous appearance, and one patient had both. The majority of lesions (87.5 %) in the two previous nD-nE groups were located in the transverse sinus and/or sigmoid sinuses (23) and cavernous sinus (5).

All seven cases of the nD-E-nS group belonged to Borden type 3 and Cognard type IIb or III (Fig. 5a, b). Thrombosis of a sinus was detected in all cases. These cases corresponded to an isolated sinus configuration (ISS), either due to thrombosis of the sinus on both sides of the draining BVs or only on one side with the other side of the sinus anatomically either disconnected from the rest of the sinus system or only connected to a BV. The 11 cases of the nD-E-S group were lesions that according to the criteria of venous drainage and direction of flow should be classified as Borden type 3 and Cognard type IV (Fig. 6a, b). One patient had ectasia, seven patients had a congestive venous appearance, and three patients had both, features that Borden classification does not take into consideration and Cognard classification records only partially. As in the previous group, they corresponded to an ISS with thrombosis detected in all cases. The majority of lesions (89 %) in the two previous nD-E (ISS) groups were located in the transverse and/or sigmoid sinuses.

The D-nE-nS and D-nE-S groups included two cases each, all Borden type 2, two Cognard type IIb located on a parietal convexial BV and on a posterior temporal BV, whereas one shunt on a posterior temporal BV was Cognard type IIa + b (Figs. 7a, b and 8a, b). For one shunt located on a superior petrosal vein, the definition of Cognard type (IIb or IIa + b) was not clear. Concerning venous strain, one lesion exhibited congestive venous appearance and the other ectatic changes.
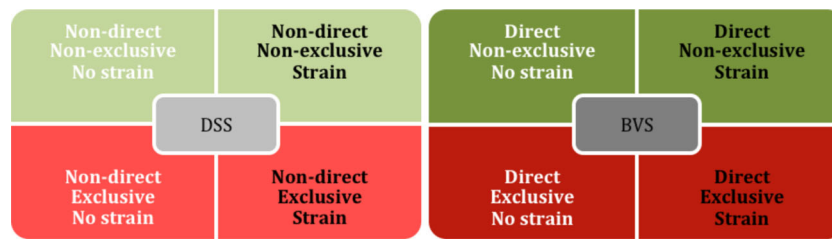
Fig. 1 The groups of the DES scheme. *The groups on the left* belong to the dural sinus shunts (DSS). *The groups on the right* belong to the bridging vein shunts (BVS). *The groups colored in tints of green* have nonexclusive LVD; therefore, they drain to both the sinus and the cortical veins; they correspond to Borden type II. *The groups colored in tints of red* have an exclusive LVD; they correspond to Borden type III. *The groups with text in white* have no signs of leptomeningeal venous strain. *The groups with text in black* have signs of leptomeningeal venous strain
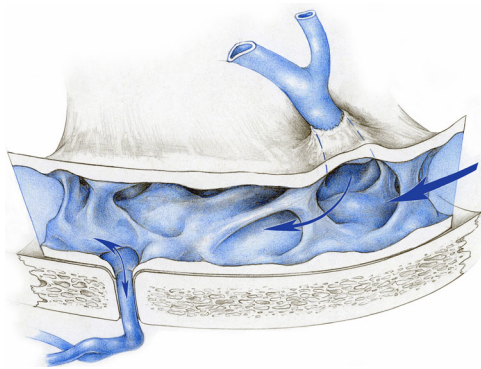

Fig. 2 Schematic illustration of an imaginary dural sinus longitudinally sectioned, with normal venous flow from right to left, a bridging vein (BV) on the right side draining into the sinus, and an emissary vein (EV) being also connected to the sinus with bidirectional flow

All 16 cases of the D-E-nS group corresponded to Borden type 3 and Cognard type III with the exception of three lesions presenting exclusive drainage to brain stem and spinal venous system (Cognard type V) (Fig. 9a, b). The majority of this group's lesions were brain stem BV shunts (nine, among them seven petrosal) and ethmoidal (5) shunts. All 37 cases of the DES group were Borden type 3 lesions, whereas 4 were Cognard type III and 33 type IV lesions (Fig. 10a, b). Two shunts had both ectasias and congestive venous appearance. The majority of this group's lesions were tentorial (12), petrosal (7), medullary BV (4), and ethmoidal (4) shunts. Superior convexial BV (3), transverse sinus BV (3), galenic (2), and sphenoid (2) shunts followed. Thrombosis of the BV exit to the sinus was seen in all cases of the above D-E groups.
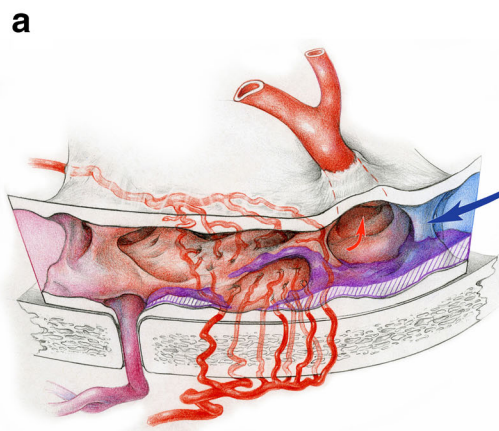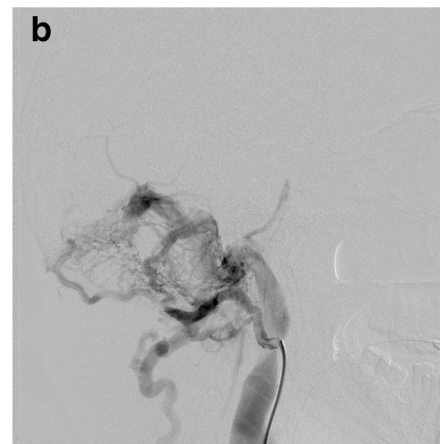

Fig. 3 a Schematic illustration of a nD-nE-nS dural shunt. The shunt is located in the sinus wall at the segment between the BV and EV. Thrombus (*colored in violet*) is also present, not necessarily obstructing the sinus. *The red curved arrow* represents the reflux to the leptomeningeal venous system through the BV. **b** An example of an extensive transverse-sigmoid sinus-based dural shunt with nondirect LVD through a temporal bridging vein to two cortical veins, which do not show any sign of strain (no strain). Actually, the drainage is mainly through the sinuses (nonexclusive LVD); anterograde flow to the jugular bulb and retrograde flow to the inferior petrosal sinus. The distal transverse sinus is thrombosed
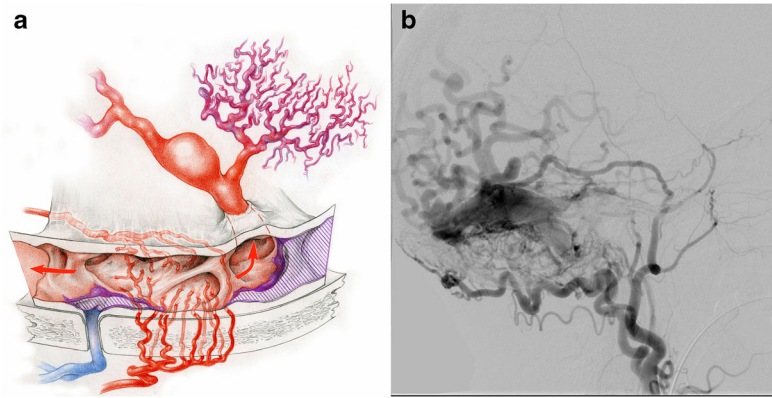
Fig. 4 **a** Schematic illustration of a nD-nE-S dural shunt. The leptomeningeal venous system displays clear signs of strain, symbolized with an ectatic change of the draining vein on the left and congestive appearance on the right. Thrombus might be, as illustrated, more extensive with obstructive phenomena. **b** An example of an extensive transverse-sigmoid sinus-based dural shunt with nondirect (nD) LVD through several temporal bridging veins to regional cortical veins. The cortical veins show clear signs of strain (S) mostly of the congestive type. The sinuses drain also the shunt (nE LVD); retrograde flow to the contralateral transverse sinus (not seen in this image). The ipsilateral distal sigmoid sinus is thrombosed
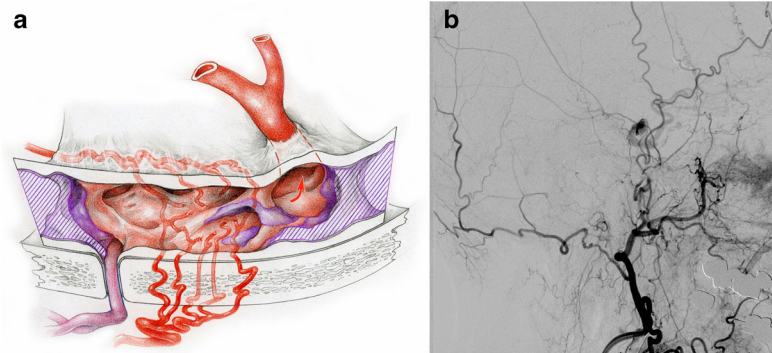

Fig. 5 **a** Schematic illustration of a nD-E-nS dural shunt. It corresponds to an isolated sinus configuration due to thrombus on either side of the BV exit. The leptomeningeal draining vein does not show signs of strain. **b** An example of a small left cavernous sinus-based dural shunt with nD, but E LVD to a contralateral orbitofrontal bridging vein, which does not show any sign of strain (nS). Apparently, the involved compartment of the cavernous sinus is isolated (likely due to thrombosis) from alternative venous exits (ISS)
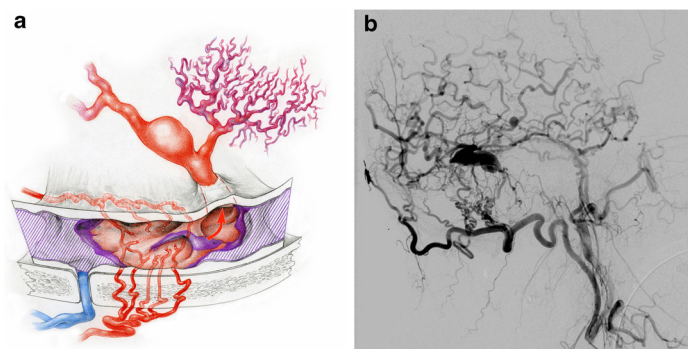

Fig. 6 **a** Schematic illustration of a nD-E-S dural shunt. In this case, the leptomeningeal vein shows clear signs of either ectasia (*left*) or congestion (*right*) or both. **b** An example of a distal transverse sinus-based dural shunt with nD, but E LVD through a Labbè bridging vein, to the whole temporal venous system with clear signs of strain (S) mostly of the congestive type. The involved compartment of the transverse sinus is isolated (ISS)
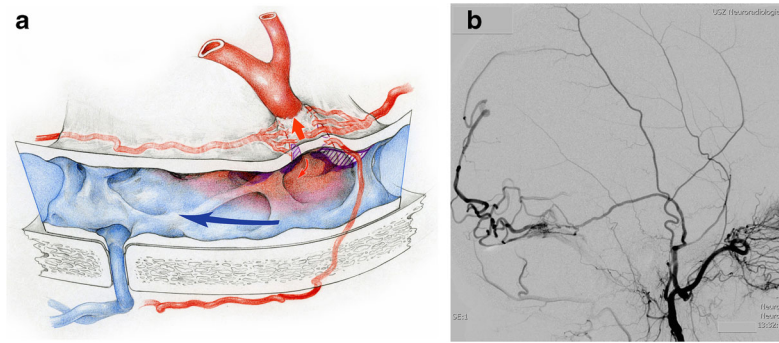
Fig. 7 a Schematic illustration of a D-nE-nS dural shunt. The shunt is located in the BV and not in the wall of the sinus; therefore, the drainage to the leptomeningeal veins is direct; it is not though exclusive. *The small curved arrow* symbolizes a small "leak" of the shunting arterial blood to the sinus. The thrombus arrangement around the BV ostium symbolizes an incomplete occlusion of the BV exit to the sinus. The leptomeningeal vein shows no signs of strain. **b** Selective injection of a left posterior temporal bridging vein-based dural shunt with D LVD into it (and secondarily to the SSS) without signs of strain (nS). The adjacent transverse sinus is almost simultaneously (nE LVD) yet faintly opacified just below the BV
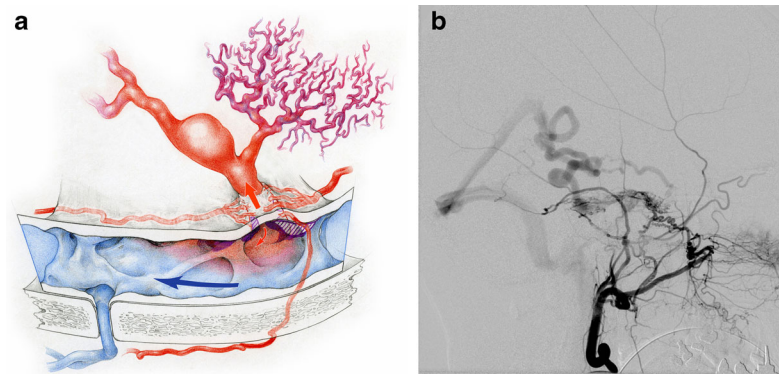

Fig. 8 a Schematic illustration of a D-nE-S dural shunt. The same arrangement as in Fig. 7, but with signs of leptomeningeal venous strain. **b** An example of a left petrosal bridging vein-based dural shunt with D LVD into it, signs of an ectatic type of strain (S) and secondary drainage to the straight sinus and contralateral transverse sinus. The ipsilateral cavernous and inferior petrosal sinus is anterograde and almost simultaneously (nE LVD) yet faintly opacified. Transvenous injections showed that the adjacent entire superior petrosal sinus is patent
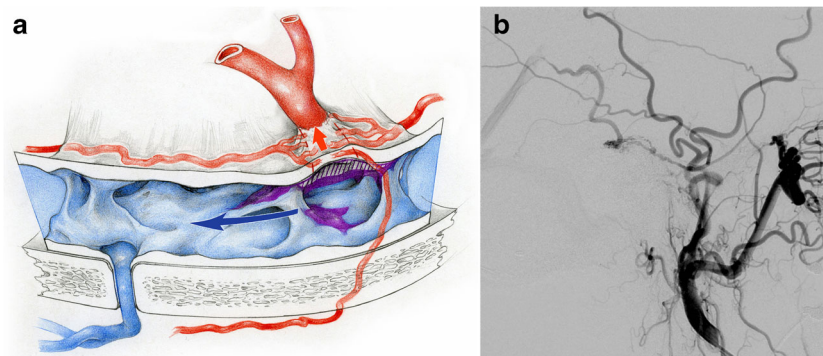

Fig. 9 a Schematic illustration of a D-E-nS dural shunt. Notice the complete occlusion of the BV exit to the sinus, which signifies an exclusive drainage to the leptomeningeal veins. The leptomeningeal veins do not show signs of strain. *The short red arrow* symbolizes a relatively low flow that may characterize these shunts in the illustrated phase. As it is shown, the BV-based shunts are infrequently supplied by transosseous feeders. **b** An example of a left petrosal bridging vein-based dural shunt with D LVD into it, without connection-reflux to the superior petrosal sinus (E LVD) and without signs of strain (nS). Secondary drainage to the straight sinus
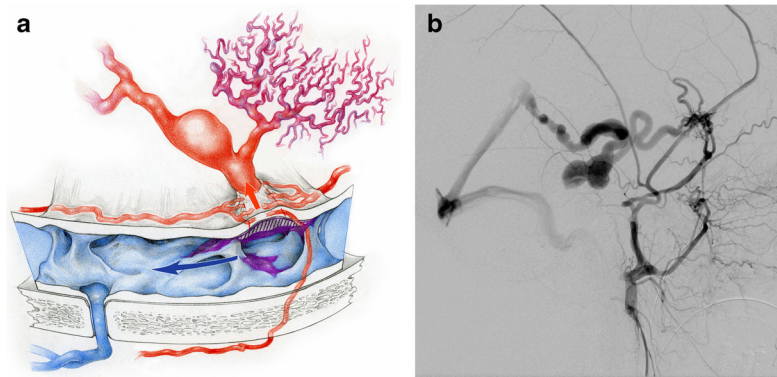
Fig. 10 **a** Schematic illustration of a D-E-S dural shunt. The same configuration as in the Fig. 9, with the exception of the leptomeningeal veins, which show clear signs of strain (more often of the ectatic type). **b** An example of a left anterior temporal bridging vein-based dural shunt (often called sphenoid wing shunt) with D LVD into it, without

connection to the sphenoparietal sinus (E LVD) and clear signs of ectatic strain (S). The anterior temporal vein empties retrograde to the ipsilateral basal vein and subsequently, through the peduncular, posterior anastomotic vein and contralateral basal vein, the shunt drains secondarily to the straight sinus

Discussion

The CDAVFs can be first classified into two basic groups: the shunts without and with LVD. The ones with LVD are prone to aggressive presentation and need further analysis. Keeping in mind that "classifications are profitable if they are accepted as incomplete and transitory from the start" [12], we will examine aspects of the currently mostly used classifications for CDAVFs.

Cognard et al. [2] defined the type III CDAVFs as those draining directly into the leptomeningeal veins, without clearly including the case of the isolated sinus due to thrombosis on both sides of the arterialized site, where the drainage into the leptomeningeal veins is indirect and which appeared as type IIb (Figs. 4 and 5 of the original publication). In the definition of the Borden [1] type III lesions, on the contrary, the case of the isolated sinus was clearly included. Despite this difference, which is evident in the original papers, subsequently, most of the publications citing the above classification systems and clarifying how they applied them both, considered the definitions of Cognard type IIb as part of Borden type II, whereas Cognard type III identical to Borden type III [8, 10, 13–16]. This discrepancy implies that comparisons between series, which do not clarify how the classification systems were meant and applied, may not be meaningful. Additionally, it highlights the potential dysfunctions and inconvenience that inevitably any conventional classification systems carry by definition. The Borden system has the major weakness that type III includes lesions with direct LVD (all the BV shunts), together with lesions displaying nondirect LVD (the case of the ISS). The Cognard classification has as confusing aspects, the poor

distinction of the presumably different types IIa and IIb (both type II) and the awkward a, b, and a + b subtypes all under the type symbolized by the number II. This inconvenience is expressed by the frequent reference to both classification systems but mostly usage of the Borden system thanks to its easy to use. Moreover, the Borden system ignores completely the very important aspect of the cerebral venous strain, expressed typically by ectatic or congested veins, whereas the Cognard system takes for granted that only type III lesions can present venous strain and only of the ectatic type (see definition of type IV).

A significant component of the Cognard system is the criterion of retrograde flow inside a sinus with or without LVD. Since sinuses have different anatomy, position, and connections, the interpretation and significance of retrograde flow into a sinus can become ambiguous. Retrograde flow into the transverse and sigmoid sinus in many cases may simply mean flow into the contralateral transverse and sigmoid sinus and further anterograde exit without obligatory involvement of the BVs. Retrograde flow into the sinus of the lesser sphenoid wing, the straight sinus, and inferior sagittal sinus may mean by definition LVD as the only exit. The difference is qualitative. The same more or less applies for the superior sagittal sinus (SSS), except for potential drainage by a parietal EV depending on the level of the SSS lesion and the patency of this EV. Retrograde flow into the cavernous sinus (CS) itself is not easily distinguishable due to its irregular shape and multiple draining routes. A CDAVF of the CS can reverse the flow into the BVs, with expected aggressive symptoms, or into the ophthalmic veins with expected orbital symptoms mostly of the benign spectrum. Retrograde flow into the superior petrosal sinus (SPS) may be connected to

either LVD into a petrosal vein or flow into the CS or both. Similarly, retrograde flow into the occipital sinus may mean either exit to the sinuses joining the torcular and further anterograde venous exit to the transverse sinuses, or retrograde flow to the SSS, or both, or exit through the occipital EV when present, or LVD through a medullary BV that may normally drain into the distal occipital or marginal sinus [17, 18]. Retrograde flow into the marginal sinus is hard to define and recognize, but theoretically, it can have an exit through hypoglossal EVs, jugular and vertebral system or LVD through a medullary BV. From the above, it becomes evident that the criterion of retrograde venous sinus flow, in terms of significance, can range from extremely significant to irrelevant. CDAVFs involving sinuses that present anatomic variations such as disconnection of the transverse sinus from the contralateral one and the SSS at the level of the torcular are equivalent to an ISS with exclusive LVD. The same applies for other sinuses, specifically the SSS, inferior sagittal sinus, sinus of the lesser sphenoid wing, and often the straight sinus, with thrombosis only downstream. This nontypical isolated sinus configuration makes the ambiguity of the above classification systems evident in the following example: a dural shunt of the sphenoid wing with distal thrombosis of the sinus (of lesser wing or sphenoparietal), retrograde flow into it and exclusive reflux into the middle cerebral vein, could be classified differently by different physicians, either as type III according to the Cognard and Borden system, accepting that the above configuration corresponds to an isolated sinus shunt, or Cognard type IIa+b taking into consideration the retrograde flow into the sinus and cortical vein. The interpretation and application of Cognard criterion of retrograde sinus flow when this flow drains exclusively into the leptomeningeal veins are not clear and may cause confusion. As a conclusion, despite the fact that retrograde flow signifies abnormal venous circulation with some degree of venous hypertension, it is still the "means" to a result that is the LVD, which is anyway recorded. Moreover, it does not reflect the capacity or incapacity of the venous system to cope, not even in the case of pure leptomeningeal retrograde flow. Then, the factors of exclusivity and directness of LVD together with potential decompensating signs of venous strain might make more sense to be recorded.

Regarding the Cognard type IV, the definition of ectasia as a venous dilatation more than 5 mm or 3 times larger than the draining vein is certainly arbitrary and problematic since the original size of the draining vein is unknown. Besides, even smaller venous aneurysms can bleed. However, the difficulty to define a size limit, above which an ectasia becomes dangerous, as well as the relativity of the ectasia definition should be considered inevitable and accepted as an inherent limitation of every analysis scheme, including ours.

Furthermore, the consideration of the spinal drainage as a qualitatively different feature, justifying a distinct, indeed higher grade, is questionable for the following four reasons. (1) In the original publication, it was stated that it was beneficial to separate out the CDAVFs with spinal venous drainage that may be responsible for myelopathy. Nevertheless, only 50 % of their cases with spinal venous drainage presented with myelopathy; 42 % presented with bleeding and 8 % with focal neurological deficit. No information about ectasias in the bleeding cases and no other justification for this different grade were provided. Subsequently, the same group of authors reported that CDAVFs of the posterior fossa with spinal venous drainage can be also asymptomatic from the spinal cord and nicely analyzed the pattern of venous drainage in relation to the cervical epidural venous exits. However, they did not consider that CDAVFs presenting without spinal-medullary symptoms had a nonexclusive spinal venous drainage in 67 % of cases, whereas CDAVFs presenting with myelopathy had an exclusive descending brain stem-spinal venous drainage in 83 % of cases [19]. (2) The fact that all these cases presented with aggressive symptoms does not make them necessarily a different or a more aggressive group if compared for instance with lesions with drainage through brain stem veins both caudally (to the spinal venous system) and cranially (to the basal system). (3) It is not clearly defined whether grade V means exclusive spinal drainage or the main drainage or just one of the draining routes; therefore, whether a shunt with nonexclusive spinal drainage with or without myelopathy should be classified as type V. (4) Since we are dealing with intracranial shunts, by definition, the spinal drainage cannot be direct. Therefore, assuming that only exclusive descending (from brain stem veins to spinal veins) spinal drainage is considered, grade V can include a wide spectrum of lesions. If a nonexclusive spinal drainage is considered, then the spectrum of lesions is even wider and the difficulties therein with the classification system even clearer.

An easy, coherent, and precise description of dural shunts, which efficiently expresses the clinically relevant anatomical and functional features of the lesion avoiding the rigidity [20] and inconvenience of some classification's awkward subdivisions, would be incomparably superior. Difficult to handle systems with ambiguous aspects are very likely to be misused [9] or equivocally applied [21].

Directness of LVD is the expression of exact location of the shunt. The BV shunts will have by definition a direct drainage to the leptomeningeal veins, whereas the sinus shunts will drain to the lumen of the sinus and then to the leptomeningeal venous system, therefore they will have an indirect LVD.

Exclusivity of LVD describes the available immediate venous exits of the shunt, therefore, is the expression of the anatomical and functional status of the involved sinuses and includes the associated outflow restrictions as complete and partial thrombosis of the sinus or stenotic phenomena regardless if they are related to thrombosis or not. Immediate venous exit means the primary, next-to-the-shunt recipient vein and

not a secondary, distant BV and sinus that comprise the ultimate, inevitably anterograde recipient of the abnormally draining leptomeningeal veins [9]. In the example of the sphenoid wing shunt used above, the lesion should be described as having exclusive LVD, though nondirect, since it is located on the sinus and not on the middle cerebral vein or other vein draining normally into the sinus.

Cortical venous strain is the expression of the venous decompensation, morphologically expressed as ectatic or congested veins (pseudophlebitic appearance), or both, determined by the previous two factors (directness and exclusivity of LVD) in combination with the venous collaterals, local venous anatomy, and variations. The strain of the venous system is not easy to evaluate, and it is even more difficult to quantify. The two aforementioned strain patterns (ectasia and congestion) are relatively straightforward to recognize and refer to qualitative features. Nevertheless, significant but otherwise uniform dilatation, without focal ectatic or aneurysmatic deformation of a draining vein, is often encountered. This, although it corresponds to a certain degree of strain, still cannot be considered a decompensation of the venous system.

Of course, the difference between a sinus that is completely occluded and a sinus that is highly stenosed downstream is qualitative, but sometimes functionally, it might be not very significant. Similarly, a slightly stenosed sinus is expected to have a less remarkable impact on the venous pressure than a highly stenosed one, both gradations not expressed in the frame of exclusivity, which is a yes-no designation. However, potential failure of the criterion of exclusivity to express the whole spectrum of alterations is presumably counterbalanced by the criterion of decompensating venous strain, which can capture the strain that violates the limits of the venous system. Otherwise, any relativity of the above definitions and potential gray zones represent the limitations of the proposed concept.

Conclusion

The classification systems, which appeared over the years and aspired to describe the features and predict the behavior of the CDAVFs, were the result of a better understanding of the disease. Whether the currently used schemes can really help in the process of further understanding and to what extent is a question without a straightforward answer. The view presented here is that they serve their purpose insufficiently. The concrete reasons supporting this view are the drawbacks of these classification systems, described in this article. Today, we can and we need to be more precise in our analysis of the lesions with LVD by describing the important and relevant aspects of their angioarchitecture in a coherent way and still easy, reproducible, and more accurate. Substantial elements of such a concept, which for brevity's sake can be called with the acronym DES, are the directness and the exclusivity of the

LVD together with the strain of the venous system. The morphological expressions of the latter and their clinical significance may need further exploration.

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Comments

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The authors proposed the so-called "DES" concept (**D**irectness and **E**xclusivity of leptomeningeal venous drainage and features of venous **S**train). This is a very practical and secure concept which is directly linked to the functional anatomy of the venous architecture of the cranium. It can be applied in clinical cases to describe the degree of parenchymal damage caused by venous hypertension associated with AV shunt at the level of dural sinuses and bridging veins.