BERVINI et al. have provided us with an excellent retrospective review of Professor Morgan’s outstanding prospective database on cerebral arteriovenous malformations (AVMs) studying the issue of transdural arterial recruitment (TDAR) to brain AVMs. I should comment that this may be the largest and most comprehensive database on a single-surgeon series of cerebral AVMs. We have learned much about these lesions from different reviews of this database for the study of different aspects of cerebral AVMs. For this particular study Dr. Morgan’s team selected 769 patients who had no history of partial treatment for their AVM and who had a complete digital subtraction angiography (DSA) assessment. Of this group 6.6% of the patients had TDAR, and the comparison of these patients with the rest of the group forms the basis of this report.

Basically, after a well-performed multivariate analysis, the factors that the authors found to have a positive statistical correlation with the presence of TDAR were as follows: increased age, increased AVM size, a combination of anterior and posterior circulation arterial supply, and, interestingly, presentation with a neurological deficit without hemorrhage. The correlation with size is not surprising. Also, it is no surprise that increasing age correlates with TDAR; most of us believe that the development of TDAR is something that occurs secondarily rather than being a primary feature of cerebral AVMs. The authors comment on that, and offer the fact that none of the very young patients in their series had TDAR as confirmation of this belief. The correlation with the presence of supply both from the anterior and posterior circulation is also not surprising, and I suspect this is a function of size—although it did come out as an independent factor in multivariate analysis. To me the most surprising correlation is that of presentation with a neurological deficit without hemorrhage, and I will comment further on this.

The main reason I agreed to comment on this paper, other than to praise the quality of the work by this group, is the fact that I have had a somewhat different, or I should rather say complementary, impression about this issue. I should acknowledge, however, that my impressions are based on simple personal experience rather than a scientific review of my personal series of AVMs, which, although significant, is less extensive than that of Professor Morgan. The factors that through the years I have come to associate with the development of TDAR are: a superficial location of the AVM, previous (in the past and not immediately preoperative) embolization, and a history of previous hemorrhage or previous surgery. That the AVM has to have a superficial location in contact with the dura mater to develop TDAR is obvious, and I am not sure why that was not one of the factors that Bervini and colleagues found to be of significance; it may simply be that they did not specifically look at this. In terms of embolization, it is a shame that Bervini and colleagues specifically excluded from analysis patients who had had previous partial embolization. It has been my contention that embolization with glue or Onyx leads to some sort of inflammatory reaction in AVMs that have a superficial location that in turn leads to the development of TDAR. I cannot prove this, but I am convinced that I see this problem much more commonly in patients who have had embolization in the past (not in the few days before surgery). In fact, I make a point of always being extremely careful opening the dura on these patients because of my certainty that they will have these difficult to deal with vascular connections between the AVM and the dura.

I have become convinced that those patients in my series who had an old hemorrhage were more likely to have TDAR. I always explained this fact to myself by thinking that something has to disrupt the normal leptomeningeal/CSF barrier between the superficial representation of the
AVM and the dura. This is also the explanation for the fact that in my experience those patients who have previously undergone operation are more likely to have TDAR, although my experience with patients who have had previous surgery for either partial removal of the AVM, feeder ligation, or any other reason is of course relatively limited. I do not have a good explanation for why in Dr. Morgan’s series a history of hemorrhage did not turn up as a significant factor for the development of TDAR. In fact, these authors specifically found that a history of neurological deficit without previous hemorrhage was a significant factor. Perhaps in their reply to this editorial they may comment on this issue.

Bervini et al. believe that the most likely explanation for the development of TDAR is that this is a secondary recruitment that results from increased shear stress in the primary arterial supply to the AVM. Their discussion in support of this theory is rather elegant, but I must confess that my simple mind is having some difficulty fully understanding it. They allude to elongation and tortuosity of the feeding arteries, which gradually brings them into contact with the dura and which then, through a process of angiogenesis and arteriogenesis, leads to the development of TDAR. I can easily visualize how these dilated arteries pulsating against the dura can produce a disruption of the normal leptomeningeal/CSF barrier and thus allow the development of TDAR. Beyond that, I admit, I cannot fully understand their proposed mechanism. Clearly, they do a very good job in their discussion in discarding other mechanisms such as one in which the TDAR is an intrinsic early feature of the malformation itself, or secondary ischemia as a result of steal into the malformation resulting in angiogenesis. I still think that inflammation brought about by embolization or disruption of the normal leptomeningeal barrier between the AVM and the dura by previous hemorrhage or surgery can be operative mechanisms in the development of TDAR.

Last, what is the importance of this phenomenon? I am convinced that the development of TDAR increases surgical difficulty, which Dr. Morgan and coauthors allude to. Interestingly, in their specific analysis of those patients from this series who had open surgery, they found no association between surgical complications and the presence of TDAR on multifactorial analysis. I suspect that this is largely due to the experience and skill of Dr. Morgan and the precautions that he takes—and describes very well in this article—when exposing cerebral AVMs with TDAR. If these precautions are not taken, catastrophe can occur from uncontrollable bleeding. I well remember a colleague’s case that occurred while I was in training. That patient exsanguinated from uncontrollable bleeding from TDAR between the tentorium and a large inferior temporal AVM. That case made a sufficient impression on me to be very vigilant ever since about the presence of this phenomenon.

In addition to the precautions during dural opening that are well described by Dr. Morgan, I frequently use preoperative embolization specifically of the dural arterial supply to avoid problems during the opening. Even with occlusion of this arterial supply the connections remain and one still has to take the same precautions to avoid tearing these connections and encountering furious arterial bleeding, which is very difficult to control either from of the dilated arterialized venous drainage or from the nidus itself during the opening. To avoid this I have frequently had to use the maneuver described by Dr. Morgan of carefully opening the dura circumferentially around the AVM and leaving a piece of dura stuck to the lesion. I would repeat, however, that this phenomenon is not common in my experience, except for those cases that have had embolization in the past, surgery, or a hemorrhage.

This is an excellent article, and I congratulate the authors on a valuable contribution.

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Response
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I am grateful for the chance to respond to the editorial review by Dr. Heros. The compliment paid to our work is very much appreciated, and I am most honored to have received this from such an outstanding surgeon and legend of cerebrovascular neurosurgery. I am not surprised that Dr. Heros found little that he did not already know in what we reported. Although what we have researched and demonstrated may be the first time that this has been reported in the literature, this work, and much of my previous work, confirmed insightful opinions, derived from thoughtful reflection on experience from such outstanding cerebrovascular neurosurgeons as Drs. Sundt, Piepgras, Heros, Drake, and Spetzler. Having had the privilege to learn from and discuss their cases with these mentors and inspirational neurosurgeons helped me choose the selection of the variables that underpin my database established in the late 1980s. The database was used to challenge some of the doctrines espoused by these neurosurgeons who expressed their personal opinions. However, more often than not, our results confirmed that these doctrines divined from personal experience were insightfully brilliant.

There is no greater or more evident example of this than the Spetzler-Martin grading system. When it was published, the scale appeared to weight variables arbitrarily in a way that was not obvious (e.g., moving from a size < 3 cm to ≥ 3 cm was equivalent to the presence of either deep venous drainage or eloquent location in a small brain AVM [bAVM]). And what may not be appreciated by those who were not involved in neurosurgery in the 1980s was the fact that not all of the variables were immediately accepted as important (i.e., deep venous drainage).
However, when we performed a multiple logistic regression on our database, incorporating a sensitivity analysis to account for those not surgically treated, we found the weighting given by Spetzler and Martin to almost perfectly reflect the weighting derived from the regression equation. This grading system proved to be a brilliant piece of inspired thinking derived from reflection on experience that has forever changed the way we think about surgery for bAVMs.

In these days, when we require such a high level of statistical examination to support opinions, we should remember that some of what might seem to be new was known and taught by these legends of cerebrovascular neurosurgery. Therefore, when Dr. Heros states that it is his “contention that embolization with glue or Onyx leads to some sort of inflammatory reaction in AVMs that have a superficial location that in turn leads to the development of TDAR…. I am convinced that I see this problem much more commonly in patients who have had embolization in the past (not in the few days before surgery),” it must be regarded as highly likely to be true. However, there are too few cases in our series of remote previous embolization for us to examine this characteristic. With regard to the statement by Dr. Heros “[t]hat the AVM has to have a superficial location in contact with the dura mater to develop TDAR is obvious, and I am not sure why that was not one of the factors that Bervini and colleagues found to be of significance; it may simply be that they did not specifically look at this.” We did not examine the juxtaposition of dura and bAVM because we deemed this a necessary precondition for the development of TDAR.

Several points for which Dr. Heros requested clarification, emphasis, or explanation follow.

1. Dr. Heros commented that he still thinks “that inflammation brought about by embolization or disruption of the normal leptomeningeal barrier between the AVM and the dura by previous hemorrhage or surgery can be operative mechanisms in the development of TDAR.” This may be true. One of the reasons that we excluded prior partial treatment was to exclude consequences of previous surgery or embolization that may have resulted in iatrogenic compromise of the normal barriers between the dura and bAVMs (and their feeding arteries). Although Dr. Heros wondered whether the 33 cases excluded in our analysis might shed light on the mechanism of TDAR, only 2 of the 9 patients with incomplete angiography who were excluded had TDAR. One of these, a 54-year-old, had multiple previous surgeries, embolization, and radiosurgery before referral. The second case, a 57-year-old had, had experienced a prior hemorrhage. Therefore, Dr. Heros may well be correct that inflammation may be a mechanism for TDAR. However, our findings suggest that this is not the only mechanism and may not be the most important explanation for the presence of TDAR. In our opinion, the presence of high wall shear stress (WSS) better fits the explanation for TDAR in our series.

2. “[A] history of neurological deficit without previous hemorrhage was a significant factor.” This fits well with our proposition that WSS is important in the development of TDAR. That high WSS has occurred in the history of a bAVM is evident from the dilated feeding arteries that are otherwise normal. However, the result of the diameter enlargement is to drive the WSS from high toward normal. In fact, findings in the limited studies examining bAVM feeding artery WSS were remarkably close to normal WSS. However, we postulate that, among patients with a neurological deficit, there are those whose neurological deficits are associated with low arterial pressures causing brain hypoperfusion. These cases have not fully compensated for the physiological demands of the bAVM, and such cases may well be associated with continued high WSS in the feeding artery. In such cases, the high WSS may drive TDAR development by angiogenesis and arteriogenesis (if opportunistic apposition to a dural surface is also present); this is a driver that is normally absent with a fully adapted feeding arterial circulation (and therefore normal WSS in the feeding artery).

3. The surgical dangers that Dr. Heros and I mention are definitely worth emphasizing. Although our results suggest that there is not an increased adverse outcome with the presence of TDAR, this came as a surprise to me because I have lost patients with catastrophic hemorrhage on opening. The reason that this does not have an impact on the overall outcome is that such complications are so dramatic, and the cause so obvious, that an example of a very small number of cases is enough to influence your surgical approach. With regard to this, it is important to emphasize a few principles. It is important to remember that a bAVM is not a tumor and that we should not think of these or approach them as tumors. What we are looking at when we see a bAVM is the normal physiological response to an arteriovenous fistula in vessels that at one stage were normal, but that have become dilated, thin-walled, and easily ruptured. This rupture can be caused by the following circumstances:

   a. exposure and tearing the vessel without prior occlusion;
   b. inappropriate occlusion technique on the thin-walled vessel; or
   c. selecting a site at which the artery proximal to the point of occlusion is too thin-walled to withstand the restoration in arterial pressure when the WSS falls to zero but the circumferential stretch increases dramatically.

The critical point that Dr. Heros and I are specifically commenting on in the presence of TDAR relates to point “a.” Trying to separate the dura from the bAVM to get to the point at which the TDAR enters the bAVM can tear the connection, with disastrous consequences. This is even worse if the TDAR is unrecognized. The safe distance from the TDAR may necessitate a wide dural window left on the bAVM from convexity dura, falx, or tentorium to eliminate tension between TDAR and dura. With respect to TDAR arising from the falx, fashioned the dural window is best approached from the contralateral side to eliminate any separation of the bAVM from TDAR. With respect to TDAR arising from the tentorium for a supratentorial bAVM, the approach may best be made at some distance from the TDAR and can be as anterior as possible.

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with division of the free edge, to eliminate the meningeal supply from the internal carotid artery, and a large dural window almost extending to the venous sinus boundaries. With respect to TDAR arising from the tentorium for an infratentorial bAVM, the approach may best be made by dividing the tentorium from a supratentorial approach to eliminate any tension between bAVM and TDAR. Further thought needs to be given to cases in which previous surgery has resulted in scalp arterial supply traversing bone and supplying the TDAR. In such cases, performing the craniotomy with multiple small strips to rapidly expose and control bleeding at each stage is critical. In my opinion, the temptation to use embolization in such cases may reduce the chances of wound healing, and I no longer consider preoperative embolization as appropriate for bAVM resection.

Finally, although Dr. Heros suggested a role for embolization of the meningeal feeders, I have not found this to be of benefit in my patients. The approach to the bAVM and dura is not altered by preoperative embolization. We appreciate the comments from Dr. Heros.

References