Multidisciplinary care of occipital arteriovenous malformations: effect on nonhemorrhagic headache, vision, and outcome in a series of 135 patients

Clinical article

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Object. In this study, the authors evaluated how an appropriate allocation of patients with occipital arteriovenous malformations (AVMs) who were treated according to different strategies would affect nonhemorrhagic headache, visual function, and hemorrhage risk levels.

Methods. Of the 712 patients with brain AVMs in the Toronto Western Hospital prospective database, 135 had occipital AVMs. The treatment decision was based on patients' characteristics, presentation, and morphology of the AVM. The management modalities were correlated with their outcomes.

Results. The mean follow-up period was 6.78 years. Nonhemorrhagic headache was the most frequent symptom (82 [61%] of 135 patients). Ninety-four patients underwent treatment with one or a combination of embolization, surgery, or radiosurgery, and 41 were simply observed. Of the 40 nontreated patients with nonhemorrhagic headache, only 12 (30%) showed improvement. In the observation group 2 patients (22%) had worsening of visual symptoms, and 2 experienced hemorrhage, for an annual hemorrhage rate of 0.7% per year; 1 patient died. In the treatment group, the improvement in nonhemorrhagic headache in 35 patients (83%) was significant (p < 0.0001). Visual deficit at presentation worsened in 2 (8%), and there were 8 new visual field deficits (9%). The visual worsening was not significantly different. There were 2 other neurological deficits (2%) and 2 deaths (2%) related to the AVM treatment. One AVM hemorrhaged. The annual hemorrhage rate was 0.1% per year. The hemorrhage risk in the observation and treatment groups was lower than the observed hemorrhage risk of all patients with AVMs (4.6%) at the authors’ institution.

Conclusions. Appropriate selection of patients with occipital AVMs for one or a combination of treatment modalities yields a significant decrease in nonhemorrhagic headache without significant visual worsening. The multidisciplinary care of occipital AVMs may aim for an apparent decrease in hemorrhage risk.

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Key Words: • arteriovenous malformation • endovascular embolization • observation • occipital location • radiosurgery • surgery • treatment • visual field

Patients with occipital AVMs commonly present with migraine-like headaches and visual disturbances.5,10 Although these symptoms seem similar, they are usually distinguishable from migraines.10,32 Because of the anatomical relationship to the visual cortex and optic radiation, the management of occipital AVMs might be associated with new VF deficits or worsening of preexisting ones. Multidisciplinary care includes endovascular embolization, microsurgical resection,2,4 stereotactic radiosurgery, and observation, depending on the patient’s age, type of presentation, extent of neurological deficit, risk of future hemorrhage,29,33 and AVM angioarchitecture.15 In this cohort, we evaluate the role of multimodality care in headache outcome, visual deficit, and hemorrhage risk in patients with occipital AVMs.
Management of occipital AVMs

Methods

Patient Population

Between September 1987 and February 2008, 712 patients were enrolled in the prospectively accrued database of Toronto Western Hospital's brain vascular malformation study group. Among them, 70 males and 65 females had occipital AVMs, and these patients were assigned to treatment or observation. The mean follow-up period was 6.78 years (range 1.5–20 years). The location of the AVM nidus was identified as medial occipital (superior and inferior calcarine region), lateral occipital (lateral to the visual radiations), anterior (anterior to the calcaine area), or the entire occipital lobe. Visual field perimetry was performed with neuroophthalmological consultation at the first visit or admission except for those patients who were admitted in a poor clinical condition. We classified the VF deficits as quadrantanopia, hemianopia, and no deficit. The VF deficit in patients who had incomplete quadrantanopia or incomplete hemianopia was approximated to the nearest quadrant. Other neurological manifestations were recorded.

Patients were added to the AVM database, and details of the angioarchitecture of the AVM Spetzler-Martin grade,26 intra- or extranidal aneurysm, venous ectasia or stenosis, pattern of venous drainage, blood supply with special attention to the calcaine artery, and the location in the occipital lobe) were evaluated.

Patients were recommended for treatment or observation after case reviews were performed by the study group. Whether treatment was necessary was based on patient age, assuming an annual 2–4% risk of bleeding.6,9 AVM presenting with hemorrhage, the architecture predisposing to higher risk of rupture (presence of intranidal aneurysms, venous stenosis, and deep venous drainage),6,9 and location and size of the AVM. The AVMs that were deemed high risk for surgery and those not indicated for radiosurgery (Spetzler-Martin Grade V and some Grade IV and those with very diffuse nidus) were assigned for observation.8

Treatment for AVMs

Treatment was intended not to cause worse or new neurological deficits unless the high risk of VF defect had been accepted by the patient and been weighed against the risk of hemorrhage. Embolization was performed as the sole technique in only a very select group of cases in which there was a significant possibility for cure. Presence of an intra- or perinidal aneurysm, and rarely, a progressive neurological deficit were among other indications. Embolization of blood supply of the AVM from the external carotid artery branches (if any) in patients suffering from severe local headache was also considered.

Patients with small AVMs (< 3 cm) who were considered for treatment were offered radiosurgery or microsurgery without embolization, given the risks and minimal benefits associated with embolization for small AVMs.17,31 Younger patients with minimal comorbidities and patients with AVM angioarchitectural risk factors for hemorrhage were preferentially treated with microsurgical resection to eliminate the risk of hemorrhage, especially when the risk associated with surgery was considered to be low.20 Older patients or those with significant comorbidities, and patients with a normal VF for whom, based on the location and the angioanatomy of the AVM, a high risk of postoperative VF deficits was estimated were preferentially treated by radiosurgery. For larger AVMs, embolization was considered to reduce the nidus size prior to surgical or radiosurgical treatment. However, we now have less enthusiasm to suggest embolization prior to radiosurgery because of a lower rate of nidus obliteration after this particular treatment combination.8 Patients’ preference was considered in equivocal situations.

For all patients who underwent surgical treatment, postoperative angiography was performed. Those in whom radiosurgery was performed underwent follow-up MR imaging every 6 months and angiography at 3 years posttreatment to confirm the AVM occlusion. Cure after radiosurgery was considered only when the nidus and the draining vein(s) both disappeared. We used these strict criteria to decrease the chance of recurrence after radiosurgery.21 Patients in whom observation had been chosen underwent clinical follow-up without new imaging unless there were new symptoms deemed related to the AVM.

Patients were followed up regularly, and their visual and neurological outcomes were assessed. All living patients underwent at least 1 formal VF test at some point during the follow-up. Nevertheless, we performed another formal VF test at the last follow-up in 112 of the 132 living patients. The VF outcome for the other 20 patients was based on the last one done during a previous follow-up. Seven reports were missing, and the gross manual VF test was considered for them.

Statistical Analysis

Statistical analysis was performed using the Fisher exact test, chi-square test, and Bonferroni-adjusted pairwise comparisons when appropriate to determine the significance of the presentation variables, AVM characteristics, and care modalities on headache; the occurrence of visual and other neurological dysfunction; and hemorrhage risk. A p value < 0.05 was considered significant.

Results

Presenting Symptoms and Findings

Patients’ characteristics including mean age and Spetzler-Martin grading in each subgroup are summarized in Table 1. In the vast majority of patients with headache, there was an absolute preference for one side of the head, ipsilateral to the AVM. The headache was described as throbbing in 44 cases and associated with visual phenomena (scintillating scotomas, blurred vision in hemifield, or flickering) in 38 cases. Visual phenomena were present in 13 cases without headache.

The frequency of dural arterial supply to the AVM in patients with headache (13 [16%] of 82) was similar to patients without headache (9 [17%] of 53) (p = 0.86).

Thirty-three patients presented with some degree of VF defect at the time of the first evaluation. Twenty-one were classified as having hemianopia and 12 quadrantan-
tanopia at the initial assessment. The frequency of VF at the first evaluation varied with the location of the AVM. Medial occipital AVMs were associated with more visual defects at presentation (p = 0.0009). The VF defect was more commonly observed in patients who experienced hemorrhage than in those with unruptured AVMs, but the difference did not reach statistical significance (p = 0.1345). Except for VF defect, the only other abnormality was papilledema in 2 patients.

Nonvisual neurological deficits were found on the initial examination in 9 patients, all of whom had experienced hemorrhage. Deficits included hemisensory abnormality, dysphasia, hemiparesis, and cognitive dysfunction. Ten patients had asymptomatic AVMs that were discovered during evaluation for other problems.

Characteristics of the AVMs

The AVM nidus ranged in diameter from 1.5 to 7 cm (mean 3.4 cm). The primary arterial supply to the AVM was through the cortical branches of the posterior cerebral artery, but all AVMs > 3 cm had multiple supplying arteries and involvement of the anterior circulation. Using the Spetzler-Martin grading system, the AVMs in this series were graded from Grades I to VI (mean Grade III).

Treatment of AVMs

Ninety-four patients (24 with a VF defect at the time of treatment) underwent one or a combination of treatment modalities based on the criteria mentioned in Methods. Glue embolization of the direct arterial supply to the AVM was performed as the sole treatment in 13 patients. Fifteen patients had embolization prior to surgery, and 13 had embolization before the radiosurgical treatment. Thirty-one patients underwent surgery alone, and 19 had only radiosurgery. Two patients had both surgery and radiosurgery, and 1 patient received all 3 treatments together (Tables 1 and 2).

Cure Rate

Thirteen patients underwent embolization alone, and the median Spetzler-Martin grade was III. Cure was the goal in 6 of these cases, and it was achieved in 4, as confirmed by the last angiogram. Other indications for embolization alone were intra- or perinidal aneurysms in 3, progressive neurological deficit in 2, and embolization of external carotid artery branches in 2 for headache control. Among a total of 46 patients who underwent resection of their AVM (median Spetzler-Martin Grade III), 41 had total removal. Residual nidi identified in 2 cases underwent reoperation, and total exclusion was confirmed. Therefore, the cure rate after surgery was 93% (43 of 46 cases). In 2 cases in which there was postsurgical residue, the patients were referred for radiosurgery. One case was cured, and the other is still in the latency period after the radiosurgery. The last surgical case with a residual nidus was lost to follow-up. Among the total of 35 patients who underwent radiosurgery (median Spetzler-Martin Grade III), cure of the AVM was achieved in 23. In 3 cases, the decision was made to perform radiation therapy again, which led to a cure in 2 (total cure 25 cases), and the remaining case has a residual micronidus without a draining vein for which cure was not considered. Further follow-up visits are scheduled to confirm the thromboses of the micronidus. In 1 case, the patient experienced hemorrhage after the radiosurgical treatment, which necessitated surgical removal of the AVM. Six patients are still in the first 3 years after the treatment; follow-up MR imaging in this subgroup has shown regression of the nidus in 4 and no change in 2. This subgroup was excluded from the final analysis of the total cure rate, and outcome as the final obliteration of the nidus is yet to be determined. In 2 patients the AVMs were not cured, and the patients did not wish further treatment. The total cure rate after radiosurgery was 86% (in 25 of 29 eligible patients). The total cure rate of multidisciplinary treatment for patients who were eligible for assessment was 82% (72 of 88).

Headache Outcome

At the last follow-up in 42 patients who had had headache before treatment, pain resolved completely in 28 (67%) and decreased > 50% in frequency in 7 (17%) (Table 3). Three patients (7%) had persistent headache despite treatment, and the remaining patients had inconsistent data. In 40 patients who had headache in the observation group, 8 (20%) improved completely over time, 4 (10%) experienced > 50% improvement, 4 (10%) worsened, 24 (60%) had no change, and 4 did not have available data. This difference was statistically significant (p < 0.0001) and confirmed that treatment yields better outcome than observation in terms of headache relief.

<table>
<thead>
<tr>
<th>Parameter</th>
<th>No. of Patients†</th>
</tr>
</thead>
<tbody>
<tr>
<td>M/F (ratio)</td>
<td>70:65 (1.07)</td>
</tr>
<tr>
<td>mean age in yrs (range)</td>
<td>36 (14–82)</td>
</tr>
<tr>
<td>mean follow-up in yrs (range)</td>
<td>6.78 (1.5–20)</td>
</tr>
<tr>
<td>HA</td>
<td>82</td>
</tr>
<tr>
<td>ruptured AVM</td>
<td>32</td>
</tr>
<tr>
<td>VF deficit at presentation</td>
<td>33‡</td>
</tr>
<tr>
<td>w/ hemorrhage</td>
<td>11</td>
</tr>
<tr>
<td>w/o hemorrhage</td>
<td>22</td>
</tr>
<tr>
<td>seizure</td>
<td>43</td>
</tr>
<tr>
<td>treatment group</td>
<td>94</td>
</tr>
<tr>
<td>treatment type</td>
<td>49</td>
</tr>
<tr>
<td>embo only</td>
<td>13</td>
</tr>
<tr>
<td>op ± embo</td>
<td>46</td>
</tr>
<tr>
<td>RS ± embo</td>
<td>32</td>
</tr>
<tr>
<td>op &amp; RS ± embo</td>
<td>3</td>
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<tr>
<td>mean age in yrs (range)</td>
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</tr>
<tr>
<td>median Spetzler-Martin grade</td>
<td>III</td>
</tr>
<tr>
<td>observation group</td>
<td>41</td>
</tr>
<tr>
<td>mean age in yrs (range)</td>
<td>57 (37–82)</td>
</tr>
<tr>
<td>median Spetzler-Martin grade</td>
<td>III</td>
</tr>
</tbody>
</table>

* † ‡ p = 0.13 for the comparison between patients with and without hemorrhage who presented with a VF deficit.
Factors Affecting Surgical Outcomes

The patients’ mean Spetzler-Martin grade was III in the surgical group. A higher Spetzler-Martin grade was not associated with more VF deficits at presentation or after treatment in the surgical group (p = 0.54). The medial occipital location was associated with more VF deficits at the initial assessment (p = 0.0009). Despite higher numbers of new or worsened VF deficits observed after surgical removal of medial occipital AVMs, the medial location was not found to be a statistically significant risk factor for surgery (p = 0.32). Patients with direct arterial supply from the calcarine branch of the posterior cerebral artery did not have a higher incidence of VF deficits at the initial assessment than those without (19 VF deficits with calcarine supply vs 14 without calcarine supply). In the surgical group, the participation of the calcarine artery was associated more frequently with new or worsened VF deficits (6 VF deficits in 20 patients with calcarine supply vs 2 VF deficits in 26 patients without calcarine supply), but the difference did not quite achieve statistical significance (p = 0.063). This is most likely due to the small number of patients with VF deficit in those subgroups.

Discussion

Headache has been reported to be present in 3–80% of patients with brain AVMs. Throbbing pain, however, with or without fortification images similar to migraines, has been reported to be more prominent and occur more often with occipital lobe AVMs. The effect of the management modality on headache can therefore influence a patient’s quality of life. In the present study, headache relief was significantly more consistent after treatment than after observation, which has been reported previously. Dural supply to the AVM was thought to be more frequent in patients presenting with headache according to Kupersmith et al., but our study did not confirm this particularity. The optimum treatment of patients

| TABLE 2: Final VF outcomes in patients who underwent treatment of their AVMs |
|----------------------------------|-----------------|-----------------|-----------------|-----------------|-----------------|
|                                | Posttreatment VF | Embo Alone       | Op ± Embo       | RS ± Embo       | Op & RS ± Embo  |
|                                | (13 patients)    | (46 patients)    | (26 patients)² | (3 patients)²   | (88 patients)²  |
| unchanged                      | 12              | 33              | 24              | 3               | 72              |
| improved                       | 0               | 5               | 1               | 0               | 6               |
| worsened                       | 1               | 8               | 1               | 0               | 10              |

* Six patients in the radiosurgery group who were still undergoing treatment were excluded from the final analysis.

Visual and Neurological Outcomes

In the subgroup of 42 patients who underwent embolization (as a sole or adjunct treatment), there was 1 new case of hemianopia due to occlusion of the calcarine artery and 1 transient hemiparesis, which fully recovered after 1 week. There were 2 fatal complications that were due to vessel rupture and hemorrhage in 1 and emboli and massive infarction in the other case.

In the surgical group, 15 patients had preoperative VF defects. Five showed improvement, 8 showed no change, and 2 had worsening of their partial defect to a complete homonymous hemianopia. There were 6 (13%) new VF deficits (4 hemianopia and 2 quadrantanopia); only 1 case of hemianopia improved to quadrantanopia at later follow-up. Therefore, the total rate of worsened preoperative visual deficit (2 of 46) or a new visual deficit after surgery (6 of 46) was 17%. One patient developed new hemiparesis and another presented with cognitive and memory problems along with Gerstmann syndrome after the surgery. No patient died.

In the radiosurgery group, 6 patients had prior VF deficits (2 quadrantanopia and 2 hemianopia), which were stable after the treatment in all except 1 in whom there was improvement from quadrantanopia to normal examination findings. There was 1 new case of hemianopia. One patient presented with significant edema, which resolved progressively over 12 months. Only 1 patient presented with hemorrhage 1 year after radiosurgery (0.5% per year), and this patient underwent microsurgical resection. No patient died. The overall bleeding rate in the treatment group was 0.1% per year.

In the observation group (median Spetzler-Martin Grade III), 2 patients experienced hemorrhage during the follow-up, yielding a hemorrhage rate of 0.7% per year. One eventually underwent surgical removal of the nidus with good outcome and the other died of hemorrhage. Of the 9 patients who presented with VF deficits in this group, 5 exhibited no change, 2 improved, and 2 worsened. Nine patients were lost to follow-up.

Statistical analysis did not reveal any significant difference in the occurrence of visual and nonvisual neurological deficits between the treatment and the observation group (p = 0.3 and p = 1, respectively). The AVM hemorrhage and mortality rates were also similar (p = 0.5 and p = 1, respectively).

Factors Affecting Surgical Outcomes

The patients’ mean Spetzler-Martin grade was III in the surgical group. A higher Spetzler-Martin grade was

<p>| TABLE 3: Outcome of headaches in both groups* |
|---------------------------------------------|---------------------------------|---------------------------------|</p>
<table>
<thead>
<tr>
<th>Parameter</th>
<th>Treatment Group</th>
<th>Observation Group</th>
</tr>
</thead>
<tbody>
<tr>
<td>total</td>
<td>42</td>
<td>40</td>
</tr>
<tr>
<td>total improvement</td>
<td>28 (67)</td>
<td>8 (20)</td>
</tr>
<tr>
<td>&gt;50% improvement</td>
<td>7 (17)</td>
<td>4 (10)</td>
</tr>
<tr>
<td>worsening</td>
<td>0</td>
<td>4 (10)</td>
</tr>
<tr>
<td>no change</td>
<td>3 (7)</td>
<td>24 (60)</td>
</tr>
</tbody>
</table>

* Data were missing in 4 patients in each group.
† The treatment group experienced a significantly better outcome than the observation group (p < 0.0001).
with AVMs located within the visual pathways should also stress protection from the risk of hemorrhage and preservation of visual function if possible.

In the treatment group, the rate of new or worsened visual deficit was 11%, new other neurological deficit was 2%, and the mortality was 2%. In the observation group, there was 22% visual worsening. Despite the trend toward more VF deficits in the treatment group and especially after surgery of occipital AVMs, there was no statistically significant difference between the observation and the treatment groups in terms of new visual deficits.

Homonymous VF loss is the most common neurological deficit in patients with occipital AVMs, occurring in 67% of cases of AVMs that have hemorrhaged compared with 25% of nonruptured AVMs. The frequency of visual loss in the present study was dependent on the exact location of the AVM nidus inside the occipital lobe with medial occipital AVMs, showing a higher rate of visual deficit at presentation. Different series have reported improvement in VF deficits ranging from 17.4 to 50% after multimodality treatment. In our series, 33 patients had preexisting VF deficits (11 with hemorrhagic origins), and 8 (24%) improved (6 patients improved after the multimodality treatment). Estimates of new or worsened VF deficits have ranged from 10 to 33% in other studies after resection. The frequency of VF loss after treatment in our series is compatible with that in previous series. Functional MR imaging has been suggested as an additional tool to better understand the cortical location of the AVM versus functional cortex and the consequence of therapy and clinical recovery.

The overall hemorrhage rates in the treatment and observation groups in the current study were 0.1 and 0.7% per year, respectively, with no significant difference. These rates were much lower than the risk of natural history of the entire cohort of patients with brain AVMs at our institution, yielding a hemorrhage rate of 4.61% per year. These rates were also lower than the most conservative yearly hemorrhage rate reported for untreated AVM in all locations. One could hypothesize that occipital AVMs might have a more benign natural history, possibly due to their more superficial location. However, there is no rationale to believe that AVMs in the occipital lobe have a lower hemorrhagic risk than other superficial locations. Furthermore, most high-risk lesions in this series were assigned to treatment and their natural history was modified; therefore, the impact of multimodality care and treatment selection cannot be ignored.

Resection of Distal Visual Pathways AVMs

In a series of 16 patients with medial occipital AVMs who had formal VF tests before and after surgery, presented with worsened VF deficits after the surgery. In a series of 23 patients reported by Bartolomei et al., 24% of patients who underwent treatment presented with new VF deficits or worsening of their preexisting deficits. Among them, 10% had permanent deficit, and 1 (4%) died. In a series of 70 patients evaluated for occipital AVMs, 46 underwent treatment. Fifteen patients (33%) had new or worsened VF deficits, 2 (4%) experienced hemorrhage, 2 (4%) died, and 4 (8%) had new neurological deficits. Sin-clair et al. recently reported a 19% rate of new VF deficit and 2% mortality. In our series and in the subgroup of patients who underwent surgery, the rate of new or worsened VF deficit was 17%.

Radiosurgery of Occipital AVMs

Although this modality is very attractive due to a very low risk of new visual problems, patients continue to be at risk for AVM hemorrhage for a period of time after radiosurgery. Even after complete obliteration, there is a very low likelihood of bleeding. The annual risk of hemorrhage is probably not affected during the radiosurgical treatment interval period. Pollock et al. reported a series of 34 patients who underwent radiosurgical treatment of their occipital AVMs. There were no formal VF evaluations, but no patient complained of new VF deficits. The obliteration rate was 71%, and the rate of bleeding during the interval period was 2.4%. The rate of new VF deficit was 3%. This shows that radiosurgery in appropriately selected patients can aim for a high obliteration rate with a low risk of bleeding (0.5% per year) and complication during the latency interval. Recently, optic radiation tractography has been integrated into radiosurgery to improve the safety of this technique and to minimize radiosurgery-induced visual disturbances. It was suggested that a maximum dose to the optic radiation of 8 Gy or more was significantly related to VF deficits or other neurological changes.

Importance of VF Defects

Patients with complete homonymous hemianopia have a poor rehabilitation outcome. Many will be unable to be gainfully employed. They generally have a disorderly visual search in their blind fields and tend to neglect the space. Some reports, however, have shown that some patients can undergo successful rehabilitation by using saccadic eye movement strategies to compensate for their VF defect, and also some VF increases have been observed at long-term follow-ups. The impact of a new VF such as a complete hemianopia should be considered as a serious disability, especially in younger patients, and be strongly considered in the management of occipital AVMs. Nevertheless, the perception of quality of life could be different in young patients who have had or are at risk of having a stroke. Shin et al. have reported that patients are less likely to take risks with their own lives than with theoretical scenarios. Therefore, despite the inherent risk of VF deficits in patients with AVMs, the clinical decision-making process should be tailored according to individual preferences.

Considerations and Recommendations

The current data represent the largest series of occipital AVMs in the literature with the longest follow-up. Patients with occipital AVMs should be evaluated by a multidisciplinary group and then be assigned to the most
Management of occipital AVMs

appropriate management arm. Although it is difficult to summarize the management strategy due to the complexity of these lesions, we provide some general recommendations; despite statistical nonsignificance, patients with AVMs in a medial occipital location or with direct calcarine supply who underwent surgery encountered a higher number of VF deficits. We therefore do not recommend surgery as the first line of treatment in this particular setting unless the high risk of hemianopia and its consequences are fully understood and accepted by the patient. This situation is best treated with radiosurgery. In selected AVMs in other occipital locations and in younger patients, surgery is the preferred treatment. High-grade occipital AVMs (Spetzler-Martin Grade V, some Grade IV, and inoperable lesions) are generally observed. Occipital AVMs without significant predictors of rupture can also be assigned to observation after careful evaluation of risks versus benefits of the treatment, especially in older individuals (> 50 years old).

Conclusions

The multimodality care of occipital AVMs with appropriate patient selection for each therapeutic arm can aim for considerable improvement in headache, without significant visual deterioration. Multidisciplinary care of occipital AVMs results in a much lower hemorrhage rate than the natural history of a general population of patients with brain AVMs.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

References


