Clinical significance of pedicle aneurysms on feeding vessels, especially those located in infratentorial arteriovenous malformations

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Object. The diminishing threshold for the application of neuroimaging leads to an increasingly frequent diagnosis of previously asymptomatic arteriovenous malformations (AVMs). In such a context, it is warranted to define the criteria that make a lesion potentially hazardous so that neurosurgeons and patients reach a decision concerning how to manage the AVM. In addition to the proposed grading system for AVMs, which assesses the risk of an actual treatment procedure, several studies have been concerned with the evaluation of angioarchitectural features. The goal of the present study is to demonstrate the significance of feeding vessel pedicle aneurysms, especially those found in infratentorial AVMs.

Methods. To determine the incidence of associated aneurysms, the authors reviewed an unselected cohort of 242 consecutive patients with AVMs managed between 1989 and 1999. Within this group were 240 patients who were treated by surgery, endovascular techniques, or a combination of both. Of these patients, 216 harbored a supratentorial and 24 an infratentorial AVM. Two additional patients with supratentorial AVMs underwent treatment of ruptured aneurysms without treatment of the AVMs. In six of the patients with supratentorial AVMs, proximal flow-related aneurysms were found on major feeding arteries, only one of which had caused hemorrhage. In only one patient were there additional distal feeding vessel pedicle aneurysms near the AVM, one of which had caused a major hemorrhage. In contrast, four of 24 patients with infratentorial AVMs had distal feeding artery pedicle aneurysms. Three of these aneurysms had caused hemorrhage.

Conclusions. Pedicle aneurysms on feeding vessels are frequently associated with hemorrhage (four of five cases in this series). In our cohort of 242 treated patients (240 treated for AVM and two for an aneurysm), feeding vessel pedicle aneurysms appear to occur more frequently in conjunction with infratentorial AVMs, which justifies aggressive management to prevent incidences of morbidity associated with rupture of the aneurysm.

KEY WORDS • cerebellar hemorrhage • interventional neuroradiology • angioma

Abbreviations used in this paper: AVM = arteriovenous malformation; DS = digital subtraction; GDC = Guglielmi detachable coil; MR = magnetic resonance; PICA = posterior inferior cerebellar artery; SCA = superior cerebellar artery.
is such a complex arrangement of pouches, fistulas, and interconnected circuitry, that these sacs might well be called aneurysms, although they bear little or no resemblance to arterial aneurysms seen in any location outside the nidus and, instead, are regular elements of this kind of pathological lesion. \(^1\)\(^4\),\(^2\)\(^8\)

In the present study, we have focused on the significance of small, distal aneurysms located close to the nidus

### Table 1

<table>
<thead>
<tr>
<th>Type of Aneurysm</th>
<th>No. of Patients</th>
<th>Location</th>
<th>Hemorrhage</th>
<th>Management of Aneurysm</th>
</tr>
</thead>
<tbody>
<tr>
<td>supratentorial AVMs resected (196 patients)</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>trunk (Type 2)</td>
<td>4</td>
<td>A2/A3</td>
<td>no</td>
<td>GDCs implanted before AVM embolization</td>
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<tr>
<td>trunk (Type 2)</td>
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<td>A2/A3</td>
<td>no</td>
<td>untreated</td>
</tr>
<tr>
<td>trunk (Type 2) &amp; pedicle (Type 3)</td>
<td>4</td>
<td>A2/A3</td>
<td>no</td>
<td>untreated</td>
</tr>
<tr>
<td>trunk (Type 2) &amp; pedicle (Type 3)</td>
<td>1</td>
<td>distal ACA</td>
<td>yes</td>
<td>resected w/ AVM</td>
</tr>
<tr>
<td>trunk (Type 2)</td>
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<td>PICA</td>
<td>yes</td>
<td>GDCs implanted, AVM untreated</td>
</tr>
<tr>
<td>infratentorial AVMs (24 patients)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>trunk (Type 2)</td>
<td>1</td>
<td>PICA</td>
<td>yes</td>
<td>only AVM removed</td>
</tr>
<tr>
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<td>distal PICA</td>
<td>yes</td>
<td>clipped</td>
</tr>
<tr>
<td>trunk (Type 2)</td>
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<td>distal PICA</td>
<td>yes</td>
<td>only AVM removed</td>
</tr>
<tr>
<td>trunk (Type 2)</td>
<td>4</td>
<td>SCA</td>
<td>no</td>
<td>Ethibloc during embolization</td>
</tr>
<tr>
<td>trunk (Type 2)</td>
<td>4</td>
<td>SCA &amp; PICA</td>
<td>yes</td>
<td>clip on SCA aneurysm</td>
</tr>
</tbody>
</table>

* Statistical analysis was performed using a Z-test for incidence of pedicle feeding artery aneurysms (one of 198 cases compared with four of 24, \(p < 0.001\)). Abbreviations: \(A2/A3\) = junction of the \(A2\) segment of the ACA and the pericallosal artery; ACA = anterior cerebral artery; MCA = middle cerebral artery.

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**Fig. 1.** Left: Computerized tomography scan of an acute intracerebral hemorrhage on the left side in the caudal aspect of the hemisphere. Right: Intraoperative view of a cerebellar AVM and a small aneurysm on the PICA at the origin of the main feeding artery to the small cerebellar AVM (arrow). The yellow discoloration of the brain adjacent to the aneurysm corresponds to a residual parenchymal hemorrhage originating from that lesion. The aneurysm was clipped during removal of the AVM.
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of the AVM. We have analyzed 242 cases of AVM that were managed in our institution over the past 10 years under a uniform protocol that calls for close neuroradiological–neurosurgical cooperation.31 We present evidence that, at least in our cohort, there appears to be a higher incidence of such aneurysms, and an association with hemorrhage, in conjunction with infratentorial AVMs. In addition to other large series,12,21 we happened to observe an unusually high incidence of these aneurysms and, therefore, can make the contribution that pedicle aneurysms on vessels feeding infratentorial AVMs exhibit features that are potentially hazardous.

Clinical Material and Methods

Patient Population

A cohort of 273 patients with cerebral AVMs has been seen at our institution since 1989. Two hundred forty were allocated to different treatment groups: surgery without previous endovascular treatment (61 patients); combination of endovascular embolization and subsequent microsurgical resection (159 patients); endovascular treatment alone (16 patients; six curative and 10 palliative); and endovascular treatment before radiation therapy (four patients). In this patient group there were 216 supratentorial and 24 infratentorial lesions. Of the 220 patients who underwent surgery, 199 were treated as elective cases and 21 as emergency cases. Two additional patients with Spetzler–Martin27 Grade V supratentorial AVMs underwent an endovascular procedure to implant GDCs to treat a ruptured aneurysm, but in both cases the AVMs remained untouched (Table 1). An additional eight patients were referred to palliative radiation therapy and 23 patients were kept under observation and their AVMs were left untreated because the estimated risks were too high or the patients refused treatment. There was a slight male prevalence in the patient population and the mean age of the patients was 33.4 years with no statistically significant difference between treated patients who had a supratentorial lesion and those who had an infratentorial one. Most patients were referred from the local population; however, some were also referred to our institution from long distances. The referral pattern has been stable over the years and a selection bias favoring complex infratentorial lesions is unlikely.

Treatment Regimen

Apart from emergency cases, the selection of a treatment regimen was made in a joint neurosurgical–neuroradiological conference, as previously described.31 It was based on risk assessment according to standard grading (Spetzler–Martin scale27), the individual patient’s vascular architecture, which was evaluated using four-vessel DS angiography, and the location of the lesion observed using standard MR imaging. In acute cases, DS angiography was performed when an atypical intracerebral hemorrhage was diagnosed in an otherwise healthy person.34 Emergency surgery was performed only in life-threatening situations; otherwise, resolution of the hemorrhage was awaited and the lesion was treated as a scheduled procedure.

Fig. 2. Computerized tomography scans (upper) and angiograms (lower). Cerebellar AVM with feeding vessels from both PICAs and also some collateral vessels from the SCA. The AVM was removed without touching the aneurysms (arrows) that had caused an intraparenchymal hemorrhage. The postoperative angiograms demonstrate complete removal of the AVM and no further filling of the feeding vessel, which carried the pedicle aneurysms.

Emboloization of the nidus was usually performed using Ethibloc.5 It has become a departmental strategy to combine embolization and surgical resection into one procedure, performed on the same day and using the same general anesthesia,33 with the intention of reducing hemorrhagic complications that could arise within the interval. The result of surgical or combined surgical–endovascular treatment was assessed by performing routine DS angiography within the first 10 days after surgical removal and regular neurological follow-up examinations.

Results

Five of 24 patients treated for infratentorial AVMs presented with aneurysms (Table 1). Four of these patients had peripheral aneurysms, of which three had bled, which was evident from the preoperative colocalization of hemorrhage and aneurysm as well as from surgical findings in which indirect signs of hemorrhage (tissue discoloration and gliosis) were seen around the aneurysms. Two of the aneurysms were located on the distal PICA and had caused parenchymal hemorrhage (Figs. 1 and 2). One patient harbored a previously ruptured aneurysm on the SCA just proximal to the AVM, which was clipped, and additional small aneurysms on collateral feeding vessels from the distal PICA, which were left untreated and appeared to have disappeared because the aneurysm-bearing segment had exclusively fed the AVM and was obliterated after removal of the AVM (Fig. 3). In another patient with an aneurysm of the SCA, the hemorrhage appeared to be distant from the aneurysm. Because the aneurysm was located within a segment exclusively feeding the AVM, it was filled with Ethibloc during embolization preceding surgical removal of the AVM (Fig. 4).
An additional patient harbored an untreated flow-related aneurysm located proximal to a cerebellar AVM. The AVM was removed and the aneurysm, which was located at the origin of the PICA from the vertebral artery and had not led to hemorrhage, was followed angiographically with indication of slow shrinking.

The situation in our cohort of patients with supratentorial lesions was somewhat different. Only five of the 196 patients treated for a supratentorial AVM had flow-related aneurysms located on major feeding arteries (Table 1). Only one of these patients had an additional distal pedicle aneurysm near the AVM with evidence of previous hemorrhage (Fig. 5).

Surgical Results

In five patients with feeding vessel pedicle aneurysms (four infratentorial and one supratentorial), the AVMs were completely surgically excised without creating a new or worsening a preexisting neurological deficit in four patients and slowly resolving aggravation of dysarthria in one patient. Proof of complete resection was obtained using DS angiography 10 days after surgery.

Management of Aneurysms

Infratentorial Pedicle Feeding Artery Aneurysms (Type 3).

One distal SCA aneurysm and one distal PICA aneurysm were clipped. One SCA aneurysm was filled with Ethibloc. Distal PICA aneurysms that were present in addition to an SCA aneurysm or were the cause of hemorrhage were left untouched and were not visible on control angiography (Table 1).

Infratentorial Main Artery Aneurysms (Type 2).

The only proximal PICA aneurysm was left untouched.

Incidence of Aneurysms and Incidence of Hemorrhage

There was a highly significant statistical difference in the occurrence of pedicle feeding vessel aneurysms in infratentorial AVMs compared with supratentorial AVMs (four of 24 compared with one of 198; p < 0.001, z-test). Likewise, the initial presentation of an acute hemorrhage, regardless of its source—aneurysm or AVM—was also significantly different between patients with infratentorial lesions and those with supratentorial ones (15 of 24 com-
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pared with 19 of 198; p < 0.001, z-test). The emphasis on acute hemorrhage in the infratentorial lesion group does not take into account that many supratentorial lesions most likely had bled previously, but had gone undiagnosed at the time of the hemorrhage because of inadequate follow up of a history of sudden headache without deficit.

Discussion

As more and more AVMs are found incidentally, there is a need for firm criteria to guide the patient and the advising neurosurgeon to make the right recommendation for appropriate management. The Spetzler–Martin grading system proposed several years ago to assess the risk of treating an AVM, based on the three parameters of size, eloquence of the affected brain region, and type of venous drainage, has already proven very useful in that respect. It does not take into account, however, angioarchitectonic features that may indicate a disproportionate risk for an individual patient to suffer neurological deficit from a hemorrhage. Aneurysms associated with AVMs, long recognized as an angioarchitectonic feature that may complicate AVM management, have been reported in approximately 5 to 7% of cases. In our series, we found flow-related extranidal aneurysms in approximately 5% of our cases, which is in good agreement with previous reports.

It has been suggested that feeding artery pedicle aneurysms, in particular, may carry an increased risk of hemorrhage. Such aneurysms present a problem that is different from intranidal aneurysms, which are a specific feature of the AVM, but may also be venous pouches and rather determine the likelihood of an AVM to bleed within itself. As stated earlier, the impact of such intranidal angioarchitectonic features is debatable without a standardized assessment and prospective evaluation, and is burdened with the difficulty of pinpointing the exact site of hemorrhage.

Exercising caution when applying statistics to small numbers, as presented in our series, our findings suggest that the risk of hemorrhage associated with pedicle aneurysms is significant because in four (80%) of five patients with such lesions evidence of previous bleeding in the anatomical location of the aneurysms was observed during surgery. In addition, by an unknown twist of fate, our series had a comparatively high incidence of infratentorial feeding artery pedicle aneurysms, which selectively allowed us to evaluate their particular significance as other studies have done, pointing out the implications of other types of aneurysms in other locations. As for the specific situation of AVMs in the infratentorial location, only recently a review focused specifically on PICA aneurysms associated with cerebellar AVMs. Similar to our cases, the source of hemorrhage in that series was almost always a distal aneurysm (16 of 18 cases specified) and the presentation was mainly that of a subarachnoid hemorrhage.

There is ongoing debate concerning when and which AVMs should be treated, and this discussion can be extended to the management of associated aneurysms. The decision of when and whether to treat an unruptured proximal AVM-associated aneurysm must be made on an individual basis, depending on the management con-
with GDCs and aneurysm discussion options with a patient. Risk assessment, which has to be taken into account when contributing just another facet to the complicated scheme of disproportionate frequency of infratentorial AVMs associated with a feeding vessel peripheral location of the aneurysm, not only in our series, but also in the literature. In that sense, our series with its higher frequency of distal pedicle aneurysms on smaller peripheral vessels feeding cerebellar AVMs in our series supports the concept that these aneurysms are flow related and are not caused by a generalized vascular pathological entity. These peripheral aneurysms are more prone to rupture than flow-related aneurysms on main trunks, which have much more stable walls. Therefore, we advocate aggressive management of these combined lesions and even suggest that an incidental cerebellar AVM that is associated with pedicle feeding artery aneurysms should be removed for the sake of eliminating the aneurysms in this potentially hazardous combination. It has to be stressed that a major risk determinant appears to be a peripheral location of the aneurysm, not only in our series, but also in the literature. In that sense, our series with its disproportional frequency of infratentorial AVMs associated with feeding artery pedicle aneurysms allows us to contribute just another facet to the complicated scheme of risk assessment, which has to be taken into account when discussing options with a patient.

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References

Fig. 6. Angiograms demonstrating an asymptomatic, proximal aneurysm (black arrow) at the A1/A2 junction, where a large callosomarginal feeding vessel for a callosal AVM originates. This AVM was treated in two steps. The first consisted of embolization of the anterior vascular territories of the AVM in the awake patient. Four weeks later the dorsal portion was embolized and the AVM was removed. Because it was anticipated that a large volume of blood flow would be diverted after obliteration of the pericallosal feeding vessel (open arrow), it was decided to close the aneurysm with GDCs (white arrow) before beginning embolization.
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