Gamma Knife surgery for intracranial aneurysms associated with arteriovenous malformations

Report of five cases

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The incidence of aneurysms coexisting with arteriovenous malformations (AVMs) ranges between 2.7 and 16.7%. The anatomical relationship between the AVM and the aneurysm is critical in deciding the best management. As a broad guide, this relationship can be classified as follows: 1) aneurysms unrelated to the AVM; or 2) aneurysms located on the feeding vessels to the nidus, which may be far away from the nidus or close to it or even within the nidus itself. Although radiosurgery has been widely accepted as a method of choice for AVM treatment, the role of radiosurgery for arterial aneurysms has not been adequately discussed.

KEY WORDS • arteriovenous malformation • aneurysm • Gamma Knife surgery • radiosurgery

From October 1994 to December 2003 the authors treated 305 patients with AVMs by using Gamma Knife surgery. Six (1.97%) of 305 patients with AVMs treated by GKS harbored associated aneurysms. Five of these patients (three men and two women) were available for a follow-up study. Three patients harbored intranidal aneurysms and two pedicular aneurysms. The mean follow-up period was 35 months. The aneurysms received a mean margin dose of 18 Gy. Over the next 36 months, the three intranidal aneurysms were completely obliterated. Of the two pedicular aneurysms, one was completely obliterated as was the AVM, and the other aneurysm disappeared but the AVM was only partially obliterated.

Radiosurgery alone may be the method of choice for treatment of an AVM with an associated intranidal aneurysm.

The incidence of aneurysms associated with AVMs that has been reported in the literature is approximately 8.9% (range 2.7–16.7%). Arteriovenous malformations associated with aneurysms appear to occur with equal frequency in both sexes.

Age has been shown to correlate positively with the presence of aneurysms. Berenstein, et al., reported that 8% of patients with AVMs who were younger than 25 years of age had concomitant aneurysms, whereas 24% of patients who were 25 to 49 years of age and 37% of patients older than 50 years of age harbored these dural lesions. Aneurysm development appears more common in association with larger AVMs with higher flow rates. Miyasaka, et al., found no associated aneurysms in patients with AVMs less than 2 cm in diameter; 13% of those patients with lesions between 2 and 5 cm in diameter harbored aneurysms, with the number rising to 37% in patients with AVMs larger than 5 cm. Brown, et al., calculated the risk of intracranial hemorrhage in the unruptured cerebral AVM at 5 years to be 7% per year in those patients with concomitant aneurysms compared with 1.7% per year for those with an AVM alone. Multiple aneurysms are more common in patients with AVMs than in the general population.

Between October 1994 and August 2004, GKS was performed in six patients with AVMs and associated aneurysms. Five patients were available for follow-up review. The mean follow up was 31 months (range 20–36 months). There were three men and two women with a mean age of 33.8 years (range 18–55 years). A summary of outcome results is provided in Table 1.

All radiosurgical procedures were performed using the Leksell Gamma Knife B unit (Elekta Instruments AB, Stockholm, Sweden). Radiosurgical dose planning was...
TABLE 1

Summary of the characteristics in five patients with concomitant aneurysms and AVMs

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs), Sex</th>
<th>Aneurysm Location</th>
<th>Follow Up (mos)</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>22, M</td>
<td>PCA</td>
<td>36</td>
<td>complete obliteration</td>
</tr>
<tr>
<td>2</td>
<td>18, M</td>
<td>rt MCA</td>
<td>36</td>
<td>complete obliteration</td>
</tr>
<tr>
<td>3</td>
<td>55, F</td>
<td>lt PICA</td>
<td>26</td>
<td>complete obliteration</td>
</tr>
<tr>
<td>4</td>
<td>55, M</td>
<td>PCA</td>
<td>23</td>
<td>complete obliteration</td>
</tr>
<tr>
<td>5</td>
<td>19, F</td>
<td>rt MCA</td>
<td>72</td>
<td>obliteration; partial AVM obliteration</td>
</tr>
</tbody>
</table>

Based on a combination of findings from stereotactic biplanar angiography and MR imaging (Gyrosan; Philips Medical Systems, Best, The Netherlands). Dose planning was performed using the KULA system (Elekta Instruments AB) until July 1997 and GammaPlan was used thereafter. The follow-up review of patients consisted of clinical examinations and MR imaging studies, which were performed at 6-month intervals after GKS. Follow-up angiography was performed when MR imaging findings revealed no residual vascular abnormality. The mean maximal dose was 36 Gy, and the mean margin dose to the AVM was 18 Gy. Of the five aneurysms, three were intranidal and two were pedicular. The mean volume of the AVM was 3.9 cm³ (range 1.6–6.5 cm³).

Case Reports

Case 1

This 22-year-old man presented with sudden onset headache and vomiting. A CT scan of the brain demonstrated spontaneous intraventricular hemorrhage and intracerebral hematoma. Angiography revealed an intranidal aneurysm of the PCA and a 2.5-cm³ AVM. On December 2, 1999, GKS was performed with a maximal dose of 35.29 Gy and a margin dose of 18 Gy to the aneurysm and AVM. Follow-up angiography and MR imaging performed 16 months later demonstrated complete obliteration of the AVM and aneurysm (Fig. 1).

Case 2

This 18-year-old man was admitted to the emergency department because of drowsiness. An initial CT scan revealed an intracerebral hematoma, and the patient underwent a craniotomy and removal of the hematoma. Postoperative angiography demonstrated a 5.77-cm³ AVM and an intranidal aneurysm fed by the right MCA and the PCA. On October 12, 1998, GKS was performed with a maximal dose of 36 Gy and a margin dose of 18 Gy. Angiography performed 36 months after GKS demonstrated complete obliteration of the AVM and aneurysm (Fig. 2).
Gamma Knife surgery for aneurysms

FIG. 2. Case 2. Neuroimages obtained in an 18-year-old man in whom the initial craniotomy was performed to evacuate an intracerebral hematoma. Postoperative angiography demonstrated an MCA aneurysm and an AVM. Gamma Knife surgery was performed to treat the aneurysm and AVM. Thirty-six months later, MR angiography demonstrated complete obliteration of the aneurysm and AVM.

Case 3
This 55-year-old woman was admitted to the emergency department because of drowsiness. An initial CT scan of the brain demonstrated spontaneous intraventricular hemorrhage, and angiography revealed three pedicular aneurysms of the left PICA and a 3.3-ml AVM. The patient refused microsurgery but did agree to undergo aneurysm and AVM embolization, which failed to obliterate either anomaly. On February 12, 2003, GKS was performed with a maximal dose of 36 Gy and a margin dose of 18 Gy. Follow-up angiography performed 25 months later revealed complete obliteration of the aneurysm and AVM (Fig. 3).

Case 4
This 55-year-old man was admitted to the emergency department because of headache and vomiting. An initial CT scan of the brain revealed spontaneous subarachnoid hemorrhage in the perimesencephalic cistern. Serial CT angiography and angiography demonstrated a right P2–3 junction aneurysm, and a P3–4 AVM with two intranidal aneurysms. Coil embolization was performed on the right P2–3 junction aneurysm and GKS was performed on the right P3–4 aneurysms and AVM. Follow-up MR angiography revealed complete obliteration of the aneurysms and AVM (Fig. 4).

Case 5
This 19-year-old woman was admitted to the emergency department because of seizures. An initial CT scan of the brain demonstrated spontaneous intracerebral hematoma in the right parietal lobe. Serial CT angiography and angiography revealed an AVM and a right intranidal aneurysm fed by the right MCA. On June 3, 1997, the patient underwent GKS performed with a maximal dose of 36 Gy and a margin dose of 18 Gy. Follow-up angiography performed 72 months after GKS demonstrated complete obliteration of the aneurysm and partial obliteration of the AVM.

Revised GKS was performed on the residual AVM.

Discussion
According to Yaşargil, Lowes and colleagues in 1925 were the first to report the occurrence of combined
FIG. 3. Case 3. Neuroimages obtained in a 55-year-old woman who harbored three left PICA pedicular aneurysms and an AVM. Embolization failed to achieve obliteration, and GKS was performed on the aneurysm and AVM. Twenty-five months postoperatively, MR angiography demonstrated complete obliteration of the AVM and aneurysms.

aneurysms and AVMs. Walsh, et al.,14 described a case of combined aneurysm and AVM. The frequency of aneurysms associated with AVMs ranges from 2.7 to 16.7%. It is important to determine the treatment options based on the anatomical location of the AVM and aneurysm. The origin of either lesion alone is unclear; however, there are three main theories that purport to explain their etiology: the coincidental theory, the congenital or developmental theory, and the hemodynamic theory. The coincidental theory was initially put forward by Boyd-Wilson;4 he suggested that associated aneurysms are simply an incidental finding. The congenital theory was developed by Arieti and Gray;1 they speculated that aneurysm formation in the setting of an AVM is a result of developmental effects. In the third theory, the hemodynamic theory, aneurysms are formed as a result of increased hemodynamic stresses placed on the walls of the major AVM feeding vessels. As with many pathogenetic theories, the truth about aneurysm development in the setting of an AVM probably cannot be explained by a single factor. Multiple factors are involved in the pathogenesis of combined AVMs and aneurysms.10

There are several classifications of aneurysms associated with AVMs.7,10,12 Perata, et al.,11 classified the association between the AVM and the aneurysm: Type 1, dysplastic or remote, unrelated to inflow vessels; Type 2, proximal, arising at the circle of Willis at the origin of a vessel supplying the AVM; Type 3, pedicular, arising from the midcourse of a feeding pedicle; and Type 4, intranidal, within the AVM nidus. Cunha, et al.,7 distinguished four categories: 1) a proximal ipsilateral major feeding artery with one subtype occurring on the contralateral side; 2) a distal superficial feeding artery; 3) a deep feeding artery; and 4) an artery unrelated to the AVM.

Redekop, et al.,12 simply categorized the aneurysm associated with an AVM as intranidal, flow-related, or unrelated to the AVM. Redekop, et al.,12 reported on 123 flow-related aneurysms in 71 patients. Eighty-four aneurysms were of the proximal type and 39 were of the distal type. Of the proximal aneurysms 26 (31%) were located on the internal carotid artery. Of the distal aneurysms 13 (33.3%) were located on the PCA. Despite ever-expanding literature on the subject of AVM-associated aneurysms, the treatment for these dural lesions remains controversial.

Cunha, et al.,7 reviewed data in 400 patients with AVMs and reported on 39 patients with 64 concomitant aneurysms. Sixty-one percent of the aneurysms in these patients were located on major feeding arteries ipsilateral to the AVM. Of the patients presenting with hemorrhage, 46% bled from the aneurysm and 33% hemorrhaged from the AVM. In 21% of patients, the source of bleeding could not be determined.

Cunha and colleagues treated the symptomatic lesion first: in 13 patients (33%) both lesions were treated at the same time. In 27 patients (69%) both lesions were completely resected. Eleven patients (28%) underwent malformation embolization, either as the only form of treatment (four patients) or as an adjuvant for surgery (seven patients). In 14 patients (36%) both lesions were treated in staged operations.

Perata, et al.,11 reported on four patients with cerebral AVMs and hemorrhage-associated aneurysms. Two
Gamma Knife surgery for aneurysms

FIG. 4. Case 4. Neuroimages obtained in a 55-year-old man. A CT scan reveals a perimesencephalic subarachnoid hemorrhage and angiography demonstrates a right P2–3 junction aneurysm and P3–4 AVMs with intranidal aneurysms. Coil embolization was performed on the right P2–3 junction aneurysm, and GKS was performed to treat the right P3–4 aneurysm and AVM. Follow-up MR angiography revealed complete obliteration of the AVM and aneurysms.

Among 217 intracerebral AVMs, Vymazal, et al., found 41 aneurysms (18.9%), which they treated with GKS. The mean age of the patients was 36.6 ± 13.9 years. The mean maximal dose was 35.4 ± 7.4 Gy, and the mean margin dose was 18.1 ± 4.1 Gy. Complete aneurysm and AVM obliteration was demonstrated in 10 patients.

In our study, GKS was performed in five AVM-associated aneurysms. The aneurysm locations were two on the MCA, two on the PCA, and one on the PICA. There were no GKS-related complications. Complete obliteration was documented in all three patients with the intranidal aneurysm–associated AVMs. Both the aneurysm and AVM in one of the two patients with pedicular

patients underwent surgery and two underwent embolization. The aneurysm was not resected in one of the two surgically treated patients when an aneurysm is located on a major, proximal, or pedicle feeding vessel (Perata Types 2 and 3), the prudent approach appears to be one of initial aneurysm obliteration, if technically feasible. For more remotely located aneurysms (Perata Type 1), if the AVM cure is dependent on radiosurgery and embolization, obliteration of the aneurysm should be performed at the first elective opportunity, as is the case with any other unruptured aneurysm (Table 2). In these cases, the risk of bleeding from the aneurysm becomes the predominant concern while waiting for the AVM to thrombose.

| TABLE 2 |
|-------------------------|------------------------|-------------------------|
| Treatment algorithm for patients with AVMs and associated aneurysms | | |
| aneurysm hemorrhage | intranidal aneurysm | treat both lesions simultaneously |
| AVM hemorrhage | intranidal aneurysm | treat both lesions simultaneously |
| | extranidal proximal aneurysm | treat aneurysm first |
| | extranidal distal aneurysm | begin AVM treatment, then |
| | | treat aneurysm |
| unknown hemorrhage | treat aneurysm first | |
aneurysms were completely obliterated and in the other patient complete obliteration of the aneurysm and partial obliteration of the AVM were achieved.

Conclusions

In patients with AVM-associated aneurysms, treatment of the hemorrhage site should be performed first or simultaneously with AVM. Gamma Knife surgery is a possible method of choice for the treatment of an AVM with an associated intranidal aneurysm.

References


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