The Association of Cerebral Aneurysms, Infundibula, and Intracranial Arteriovenous Malformations

KAZUO MIYASAKA, SAMUEL M. WOLPERT, AND ROBERT J. PRAGER

SUMMARY Intracranial arteriovenous malformations (AVMs) are congenital communications between the cerebral arteries and veins without an intervening capillary network.1-3 The association of the malformations with other vascular lesions such as intracranial aneurysms has previously been reported.4-18 We have reviewed 132 consecutive patients with intracranial AVMs evaluated angiographically at New England Medical Center. In addition to the expected high incidence of saccular aneurysms, a high incidence of arterial infundibula was found. A review of the association between the AVMs, aneurysms and infundibula, and possible etiologic factors in the relationship forms the subject of this report.

Patients and Methods

The angiograms of 132 consecutive patients with intracranial AVMs were reviewed and the incidence of associated aneurysms and infundibula studied. There were 65 males and 67 females, ranging in age from 6 to 71 years. [Infundibula are generally considered to be conical outpouchings, no greater than 3 mm in diameter, located at arterial origins, with small arteries originating from their apices.17-19 We considered that restricting infundibula to a maximal diameter no greater than 3 mm at the base is arbitrary. In all the infundibula larger than 3 mm, small arterial branches originated from the apices; in the smaller infundibula, arterial branches originated from the apices in most cases.] No aneurysms were found in the 30 patients with small AVMs. Aneurysms were encountered in 9 of the 67 patients with moderate-sized AVMs and in 13 of the 35 patients with large AVMs. The distribution of AVM sizes in the patients with aneurysms was statistically different from that in patients without aneurysms ($x^2 = 12.3, p < .001$), indicating that aneurysms were more likely in the patients with larger AVMs.

Results

1. Saccular aneurysms

Forty-three intracranial aneurysms were found in 22 of the 132 patients (16.7%) (table 1). Thirteen patients had single aneurysms only, three patients had 2 aneurysms each, four patients had 3 aneurysms each, and two patients had 5 and 7 aneurysms respectively. There were 12 females and 10 males.

The ages of the patients, the sizes of the AVMs, and the sites of the aneurysms were evaluated.

a) Age

The mean age of the 22 patients with aneurysms was $41 \pm 12.9$ years (range: 21-68 years), where that of the 110 patients without aneurysms was $31 \pm 12.7$ years (range: 6-73 years). The difference was statistically significant ($p < .01$) indicating that aneurysms were more likely to occur in the older patients.

b) Size of the AVMs (Table 2)

The AVMs were arbitrarily divided into three groups: small ($2 \times 2 \times 2$ cm or less, 30 patients), moderate ($2 \times 2 \times 2-5 \times 5 \times 5$ cm, 67 patients), and large ($5 \times 5 \times 5$ cm or larger, 35 patients). Because the demarcation between the nidus of the malformation and the feeding arteries and drainage veins was often not precise, these measurements were often "guessmates." [The measurements given here (and for the infundibula) are those obtained after correcting for magnification factors of 1.1 on nonmagnified lateral films, 1.2 on nonmagnified AP films, and 1.8 on magnified lateral films. These factors are for a midcranial structure such as the pineal gland. Corrections for the position of the AVMs within the cranium were not made.] No aneurysms were found in the 30 patients with small AVMs. Aneurysms were encountered in 9 of the 67 patients with moderate-sized AVMs and in 13 of the 35 patients with large AVMs. The distribution of AVM sizes in the patients with aneurysms was statistically different from that in patients without aneurysms ($x^2 = 12.3, p < .001$), indicating that aneurysms were more likely in the patients with larger AVMs.

c) Site of the Aneurysms (Table 3)

There was no predilection for the aneurysms to occur on the Circle of Willis. Sixteen of the 43 aneurysms (37.2%) occurred on the internal carotid artery, with 10 aneurysms (23.2%) on the carotid siphon (fig. 1) and 6 (15%) on the terminal internal carotid artery. Six of 43 aneurysms (14.0%) were located on the anterior cerebral artery, and 7 (16.3%) were on the middle cerebral artery. Four of the 13 anterior and middle cerebral artery aneurysms were located on branches distal to the anterior communicating artery or distal to the knee of the middle cerebral artery (fig. 2).

Fourteen of the 43 aneurysms (32.6%) originated from the arteries of the vertebrobasilar system. Six were on the posterior cerebral artery in its peduncular segment (fig. 3), 3 were on the basilar artery either at the apex or just below the superior cerebellar artery (fig. 4), 3 were on the basilar artery between the posterior cerebral and superior cerebellar arteries (fig. 4), and 2 arose from the anterior inferior cerebellar artery, at its origin from the basilar artery in one case and from distal branches of the anterior inferior cerebellar artery in another (fig. 5). Nine patients had multiple aneurysms; in six of these patients, the aneurysms were on the same artery and were juxtaposed to each other (figs. 4 and 5).

The relationship between the site of the aneurysms
and the feeding arteries to the AVMs was explored. Eighteen of the 43 aneurysms were located on the intracranial arteries specifically supplying the AVM (figs. 1–5). Sixteen (37.2%) were on the internal carotid artery ipsilateral to the supratentorial AVMs, or on the basilar artery with posterior circulation AVMs (figs. 1, 4). Seven of the remaining 9 aneurysms originated from arteries of the contralateral carotid.

**Figure 1.** Parietal lobe AVM. Right carotid angiogram, frontal (A) and lateral (B) projections. Two lobulated aneurysms originate from the carotid siphon near the origin of the ophthalmic artery. Three aneurysms are located on the middle cerebral artery, on the M1 segment and at the knee. Infundibula are seen on the internal carotid artery, one (arrow) at the origin of the posterior communicating artery and one (arrow) at the origin of the anterior cerebral artery. Note that the A1 segment arises from the apex of the infundibulum.
siphon or internal carotid/posterior communicating artery junctions. In these 7 cases, the contralateral carotid artery contributed to the AVM through the anterior communicating artery. Two aneurysms were not apparently related to the AVM. Thus, 41 of 43 aneurysms (95%) appeared to be related anatomically.

TABLE 1. Incidence of Aneurysms and Infundibula in 132 Patients With Intracranial Arteriovenous Malformations

<table>
<thead>
<tr>
<th></th>
<th>No. of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>Aneurysms alone</td>
<td>12</td>
</tr>
<tr>
<td>Aneurysms and infundibula</td>
<td>10</td>
</tr>
<tr>
<td>Infundibula alone</td>
<td>20</td>
</tr>
<tr>
<td>Total</td>
<td>42</td>
</tr>
</tbody>
</table>

Incidence of aneurysms 22/132 = 17%.
Incidence of infundibula 30/132 = 23%.

TABLE 2. Relationship of Size of AVMs to Aneurysms and Infundibula

<table>
<thead>
<tr>
<th>Size of AVM</th>
<th>No. of patients</th>
<th>No. of patients with aneurysms</th>
<th>No. of patients with infundibula</th>
</tr>
</thead>
<tbody>
<tr>
<td>Small</td>
<td>30</td>
<td>0</td>
<td>4</td>
</tr>
<tr>
<td>Moderate</td>
<td>67</td>
<td>9</td>
<td>13</td>
</tr>
<tr>
<td>Large</td>
<td>35</td>
<td>13</td>
<td>13</td>
</tr>
<tr>
<td>Total</td>
<td>132</td>
<td>22</td>
<td>30</td>
</tr>
</tbody>
</table>

TABLE 3. Site of Aneurysms and Infundibula

<table>
<thead>
<tr>
<th>Parent artery</th>
<th>No. of aneurysms</th>
<th>No. of infundibula</th>
</tr>
</thead>
<tbody>
<tr>
<td>ICA</td>
<td></td>
<td></td>
</tr>
<tr>
<td>intracavernous</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>ophthalmic origin</td>
<td>9</td>
<td>2</td>
</tr>
<tr>
<td>posterior communicating</td>
<td>4</td>
<td>22</td>
</tr>
<tr>
<td>anterior choroidal</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>carotid bifurcation</td>
<td>1</td>
<td>16</td>
</tr>
<tr>
<td>ACA</td>
<td></td>
<td></td>
</tr>
<tr>
<td>A. comm. or AI</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>distal</td>
<td>2</td>
<td>6</td>
</tr>
<tr>
<td>MCA</td>
<td></td>
<td></td>
</tr>
<tr>
<td>proximal to knee</td>
<td>5</td>
<td>4</td>
</tr>
<tr>
<td>distal</td>
<td>2</td>
<td>7</td>
</tr>
<tr>
<td>PCA</td>
<td>6</td>
<td>0</td>
</tr>
<tr>
<td>Basilar</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Apex</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>PCA/SCA junction</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>upper basilar</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>AICA</td>
<td>2</td>
<td>8</td>
</tr>
<tr>
<td>Total</td>
<td>43</td>
<td>34</td>
</tr>
</tbody>
</table>

ICA = internal carotid artery; ACA = anterior cerebral artery; MCA = middle cerebral artery; PCA = posterior cerebral artery; SCA = superior cerebellar artery; AICA = anterior inferior cerebellar artery; AI = horizontal portion of the anterior cerebral artery; A. comm = anterior communicating artery.

FIGURE 2. Parietal lobe AVM. Left carotid angiogram, frontal (A) and lateral (B) projections show an aneurysm (arrow) located on the pericallosal artery.
FIGURE 4. Occipital lobe AVM. Left vertebral angiogram demonstrates three saccular aneurysms arising from the basilar artery: one at its apex; another at the junction between the posterior cerebral and superior cerebellar arteries; and a third from its mid-portion. This latter aneurysm was confirmed at surgery to have bled.

or hemodynamically to the arteries supplying the AVMs.

2. Infundibula

Thirty-four infundibula were present in 30 of the 132 patients (22.7%) (table 1). The bases of the infundibula were 3 mm or greater in 16 patients (table 4); in 8 of these latter patients, saccular aneurysms coexisted. In 14 patients, the infundibula were less than 3 mm in diameter; in only two patients were coexisting aneurysms present. Statistically, this difference is significant, indicating a relationship between the size of the infundibula and the presence of aneurysms ($x^2 = 5.4$, $p < .025$).

The ages of the patients, the sizes of the AVMs, and the sites of the infundibula were evaluated.

<table>
<thead>
<tr>
<th>Size of infundibula</th>
<th>No. of patients</th>
<th>Presence of aneurysms</th>
</tr>
</thead>
<tbody>
<tr>
<td>3 mm or greater</td>
<td>16</td>
<td>8</td>
</tr>
<tr>
<td>less than 3 mm</td>
<td>14</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>30</td>
<td>10</td>
</tr>
</tbody>
</table>

$x^2 = 5.4$, $p < 0.025$. 

TABLE 4. Size of Infundibula Related to Coexistent Aneurysms
FIGURE 5. Fourth ventricular AVM. Left vertebral angiogram, Caldwell view. (A) There is a questionable aneurysm (arrow), diagnosed retrospectively, on the right anterior inferior cerebellar artery. (B) Six years later, an unequivocal aneurysm (arrow) is seen on the right anterior inferior cerebellar artery.

a) Age

The mean age of the 30 patients with infundibula was 39 ± 12.6 years (range: 13–68 years), whereas that of the 92 patients without infundibula was 30 ± 13.1 years (range: 6–73 years). The difference was statistically significant (p < .01), indicating that infundibula were more likely to occur in the older patients.

b) Size of AVMs (Table 2)

Infundibula were found in 4 of the 30 patients with small AVMs, 13 of 67 patients with moderate-sized AVMs, and in 13 of the 35 patients with large AVMs. The distribution of AVM sizes in the patients with infundibula was statistically different from that in the patients without infundibula (χ² = 6.1, p < .05), indicating that infundibula were more likely in the patients with larger AVMs.

c) Site of the Infundibula (Table 3)

Most of the infundibula occurred on the internal carotid artery at its junction with the posterior communicating artery (22 of 34 infundibula, 67.4%) (figs. 1, 6). In 2 patients, the infundibula occurred at the origin of the ophthalmic artery, and, in one patient, an infundibulum was present at the origin of the anterior choroidal artery. Nine of the infundibula were located on the middle or anterior cerebral arteries (figs. 1, 6). None of the infundibula occurred on the posterior circulation. In none of the patients with multiple infundibula were the infundibula in juxtaposition with each other.

Twenty-eight of the 34 infundibula (92%) (table 5) were located either on arteries supplying the AVMs or on an internal carotid artery ipsilateral to the AVMs. Six infundibula were located on the internal carotid artery contralateral to the AVM; however, in none of these did the contralateral carotid artery supply the AVM.

Figure 6. Parietal lobe AVM. Right carotid angiogram. (A) Two infundibula measuring greater than 3 mm in diameter arise from the internal carotid artery: at the origin of the posterior communicating artery (open arrows); and from the middle cerebral artery at the origin of the insular branch (arrow). (B) After surgical removal of the AVM, one of the internal carotid infundibula has decreased in size together with reduction in size of the internal carotid artery. The insular branch infundibulum has not changed in size.
3. Changes in Size of Saccular Aneurysms and Infundibula With Therapy (Table 6)

In 13 of the 22 patients with aneurysms, artificial embolization of the AVM was the initial treatment. None of the aneurysms changed in size as assessed on the immediate postembolization angiograms. In 4 patients, excision of the AVMs was the prime therapy; in 3 patients, the aneurysms decreased in size after surgery (fig. 3). In one patient whose AVM was not treated, an aneurysm which was not initially identified was seen 6 years later on a follow-up angiogram (fig. 5).

In 4 patients, the infundibula decreased in size after treatment of the AVMs (2 patients following embolization, and 2 following surgery).

**Discussion**

The association of saccular aneurysms with intracranial AVMs, whether they represent related congenital anomalies or whether the AVMs antecede the development of the aneurysms, has been a controversial subject for many years. The association has often been reported, with the incidence varying between 2.7% and 9.3%. The hypothesis that both conditions are based on the same developmental defects is unlikely since, in an autopsy study on 215 subjects with cerebral aneurysms, the prevalence of AVMs was not significantly higher than in a control group of 849 subjects without cerebral aneurysms. Another possibility is that aneurysms and AVMs occur coincidentally. This association of rare lesions, though possible, is considered to be unlikely.

The present and other studies suggest that hemodynamic stresses due to increased arterial flow to the aneurysms are probably major factors in their etiology. The aneurysms in our series occurred at sites remote from the Circle of Willis, and were commonly located on the arteries contributing to the AVMs (figs. 2–6). Two or more aneurysms were often juxtaposed on the same artery (figs. 2, 5).

Experimentally, increased blood flow can cause distension and degeneration of the arterial walls. In a postmortem study of a case with an AVM and aneurysm, Brihaye and Blackwood found that the media of the dilated artery varied in thickness and was composed of fibrous and elastic tissue, and the elastic lamina was interrupted in some places. Shenkin et al described 6 patients with intradural carotid aneurysms treated by carotid ligation. In 2 of the 6 patients, angiography carried out 3–10 years later demonstrated aneurysms that had developed on the opposite internal carotid artery. Hemodynamically, a similar situation can occur in the cases where the AVMs are supplied by the contralateral internal carotid artery.

Infundibula were found associated with the AVMs in 30 patients. To the best of our knowledge, this is a new observation. The incidence of 22.7% is significantly greater than the 6.6% incidence found in an angiographic study of 1020 normal patients (x^2 = 38.0, p < .001), but not significantly greater than the 17.7% reported incidence in 291 brains studied with postmortem angiography.

The infundibula are located on the feeding arteries to the AVMs, and occur not only on the internal carotid artery but also on the branches of the anterior and middle cerebral arteries (fig. 6), again suggesting an augmented cerebral blood flow in their etiology. In one case in this series, a large infundibulum measuring 3.5 mm in diameter before therapy decreased to 2.0 mm in diameter after artificial embolization, and again increased to 3.0 mm in diameter as the caliber of the parent artery changed. Further support for the hemodynamic theory is the fact that surgical treatment of the AVMs, which in some cases results in a decrease in size of the feeding arteries, is followed by shrinking of the aneurysms and infundibula (figs. 3, 6).

In our series of 132 patients, 6 presented with intracranial hemorrhages. In 3 patients, two with aneurysms on the peduncular segment of the posterior cerebral artery and one with a basilar artery aneurysm immediately below the superior cerebellar artery, it was surgically confirmed that rupture of the aneurysms had caused the hemorrhages. It has been previously observed that, after surgery or artificial embolization of AVMs, the feeding arteries and their main branches decrease in size while the adjacent normal arteries or adjacent feeding arteries increase in size. These changes suggest that treatment of the AVM per se may possibly protect the patient from rupture of aneurysms located on the feeding artery. Conversely, when aneurysms are located on adjacent feeding arteries to an AVM or arteries adjacent to

<table>
<thead>
<tr>
<th>TABLE 5. Relationship of Size of Infundibula to Feeding Arteries</th>
</tr>
</thead>
<tbody>
<tr>
<td>Size of infundibula</td>
</tr>
<tr>
<td>---------------------</td>
</tr>
<tr>
<td>3 mm or greater</td>
</tr>
<tr>
<td>less than 3 mm</td>
</tr>
<tr>
<td>Total</td>
</tr>
</tbody>
</table>

\[ \chi^2 = 0.8, p < 0.1. \]

<table>
<thead>
<tr>
<th>TABLE 6. Changes in Size of Aneurysms and Infundibula With Therapy</th>
</tr>
</thead>
<tbody>
<tr>
<td>Aneurysms</td>
</tr>
<tr>
<td>-----------</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Aneurysms</td>
</tr>
<tr>
<td>Infundibula</td>
</tr>
</tbody>
</table>

*In one patient, both embolization and removal of the AVM caused a decrease in size.
but not supplying an AVM, it may be advisable to
treat the aneurysms directly before treating the AVMs
by embolization.

Conclusions

Saccular aneurysms and infundibular widenings are
frequently associated with AVMs. The incidence of
aneurysms correlated with increasing patient's age
and with the size of the AVMs. Both saccular aneu-
rysms and infundibula are usually located on the
arteries supplying the AVMs. The carotid siphon, the
distal branches of the anterior and middle cerebral
arteries, and the arteries of the basilar system are
often sites for the aneurysms and the infundibula. Two
or more aneurysms often occur on the same artery in
juxtaposition to each other. Some aneurysms and in-
fundibula decrease in size after treatment of the
AVMs. The results confirm the high incidence of in-
fundibula and aneurysms with AVMs, and support the
concept that increased blood flow may be a major
cause of this association.

References

more: Williams & Wilkins, 1977: 127-141
3. Walsh FB, King AB: Ocular sign of intracranial saccular aneu-
rysms: Experimental work on collateral circulation through the
ophthalmic artery. Arch Ophthal (Chicago) 27:1-33, 1942
4. Arieti S, Gray EW: Progressive multiform angiosis. Associa-
tion of a cerebral angiomata, aneurysms and other vascular
changes in the brain. Arch Neurol Psychiat 51: 182-189, 1944
5. Anderson RMcD, Blackwood W: The association of arterio-
venous angiomata and saccular aneurysm of the arteries of the
6. Patterson JM, McKissock W: A clinical survey of intracranial angiomata with special reference to their mode of progression
and surgical treatment; A report of 110 cases. Brain 79: 233-266, 1956
7. Brihaye J, Blackwood W: Arterio-venous aneurysm of the cere-
8. Boyd-Wilson JS: The association of cerebral angiomas with in-
9. Reigh EE, Lemmen LJ: Cerebral aneurysms with other intra-
10. Cronqvist S, Troopp H: Intracranial arteriovenous malforma-
tions and arterial aneurysms in the same patient. Acta Neurol
cranial aneurysms and subarachnoid hemorrhage. Section VI.
Arteriovenous malformations. An analysis of 545 cases of cranic-cerebral arteriovenous malformations and fistulae
reported to the cooperative study. J Neurosurg 25: 467-490,
12. Shenkin HA, Jenkins F, Kim K: Arteriovenous anomaly of the
brain associated with cerebral aneurysm. Case report. J
Neurosurg 34: 225-228, 1971
malformation studied by angiography: Multiple aneurysms
angiomas and arterial ectasia. Neuroradiology 5: 117-123,
1973
14. Suzuki J, Onuma T: Intracranial aneurysms associated with
15. Higashi K, Hatano M, Yamashita T, Inoue S, Matsumura T:
Coexistence of posterior inferior cerebellar artery aneurysm
and arteriovenous malformation fed by the same artery. Surg
Neur 12: 405-408, 1979
16. Koulouris S, Rizzoli HV: Coexisting intracranial aneurysm and
17. Saltzman GF: Infundibular widening of the posterior com-
municating artery studied by carotid angiography. Acta Radiol
51: 415-421, 1959
18. Hassler O, Saltzman GF: Histologic changes in infundibular
widening of the posterior communicating artery. A prelimi-
19. Taveras JM, Wood EH: Diagnostic neuroradiology. Baltimore:
Williams & Wilkins, 1976: 585
Diagnostic neuroradiology. St. Louis: Mosby, 1974: 2355-2489
21. Wollschlaegar G, Wollschlaeger PB, Lucas FV, Lopez VF: Ex-
municating artery studied by carotid angiography. Acta Radiol
51: 415-421, 1959
22. Wolpert SM, Stein BM: Catheter embolization of intracranial
Louis: Mosby, 1972: 351-470
24. Roach MR: Changes in arterial distensibility as a cause of post-
stenotic dilatation. Amer J Cardiol 12: 802-815, 1963
25. Fry DL: Acute vascular endothelial changes associated with in-
26. Stehbens WE: Blood vessel changes in chronic experimental
The association of cerebral aneurysms, infundibula, and intracranial arteriovenous malformations.

K Miyasaka, S M Wolpert and R J Prager

Stroke. 1982;13:196-203
doi: 10.1161/01.STR.13.2.196

Stroke is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
Copyright © 1982 American Heart Association, Inc. All rights reserved.
Print ISSN: 0039-2499. Online ISSN: 1524-4628

The online version of this article, along with updated information and services, is located on the World Wide Web at:
http://stroke.ahajournals.org/content/13/2/196

Permissions: Requests for permissions to reproduce figures, tables, or portions of articles originally published in Stroke can be obtained via RightsLink, a service of the Copyright Clearance Center, not the Editorial Office. Once the online version of the published article for which permission is being requested is located, click Request Permissions in the middle column of the Web page under Services. Further information about this process is available in the Permissions and Rights Question and Answer document.

Reprints: Information about reprints can be found online at:
http://www.lww.com/reprints

Subscriptions: Information about subscribing to Stroke is online at:
http://stroke.ahajournals.org//subscriptions/