Intracranial aneurysms associated with arteriovenous malformations

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Nine cases of intracranial aneurysm associated with cerebral arteriovenous malformation (AVM) were found in a total of 140 patients with cerebral AVM (6.4%). Radical operation was performed for both angioma and aneurysm in eight cases and for aneurysm only in one case. The suitability of surgical treatment of both aneurysm and angioma is discussed.

KEY WORDS • aneurysm • arteriovenous malformation

Cases of cerebral aneurysm associated with arteriovenous malformation (AVM) are fairly rare, and questions remain with regard to etiology and treatment. Between 1959 and 1975, we have treated 140 cases of AVM (excluding cases of dural AVM), among which nine patients were found to harbor cerebral aneurysms as well. All but one patient with a deep-seated AVM were cured by total extirpation of the AVM during a radical operation on the aneurysm. During surgery, 500 to 1000 cc of 20% mannitol solution was administered intravenously under normotensive and normothermic anesthesia, to prolong the time during which the cerebral arteries could be occluded.

We report these cases, and briefly discuss surgery on this type of lesion.

Summary of Cases

Of the nine patients, four were men and five were women. Onset of symptoms occurred between the ages of 21 and 52 years, with an average age of 38 years. Compared with the 27-year average found at our Clinic for AVM cases, this is an advanced age, but it is quite young in comparison with cerebral aneurysm cases in general. The initial symptom was hemorrhage in six patients, two of whom developed intracerebral hematomas and hemiparesis, and three of whom developed convulsive seizures. One of these latter cases had a hemorrhage 10 years after onset of convulsive seizures. In all nine cases the aneurysms were found on the same side as the main artery feeding the AVM's.

In three cases the aneurysms were located at the junction of the internal carotid and posterior communicating arteries, in three cases on the pericallosal branch of the anterior cerebral artery, in one case at the internal carotid artery bifurcation, in one case on the peripheral branch of the middle cerebral artery, and in one case on the posterior inferior cerebellar artery. No cases of multiple aneurysms were found (Table 1). Typical angiograms clearly demonstrating the aneurysm and the AVM are presented in Figs. 1, 2, and 3.

Discussion

Arteriovenous malformations associated with cerebral aneurysms are relatively rare. Before the widespread use of angiography, such lesions were discovered only by chance at autopsy. Moniz and Guerra, Laur, and others reported angiography of this anomaly, and its diagnosis is now possible. In 1958, Boyd-Wilson reported 16 cases in the literature, and thereafter case reports have appeared sporadically. Paterson and McKissock reported that the incidence of this complication was 2.7%; that is, out of 110 cases of AVM, they found three with associated aneurysms. Cronqvist and Troupp reported 13 cases of aneurysms in 150 AVM cases (8.7%), and Perret and Nishioka reported 37 cases in 490 AVM cases (7.6%) in the Cooperative Study. We found a rate of 6.4% in our series. Anderson and Blackwood reported an autopsy series with a high incidence of aneurysms associated with AVM's, namely,
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TABLE 1
Association of cerebral AVM and intracranial aneurysm

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Sex, Age (yrs)</th>
<th>Symptoms</th>
<th>Location</th>
<th>AVM</th>
<th>Operation</th>
<th>Aneurysm</th>
<th>AVM</th>
<th>Outcome†</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M, 39</td>
<td>convulsion</td>
<td>rt ACA</td>
<td>rt parieto-occipital</td>
<td>ligation</td>
<td>extirpation</td>
<td>A</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>F, 47</td>
<td>SAH</td>
<td>rt ACA-PCoA junction†</td>
<td>rt frontal†</td>
<td>clipping</td>
<td>extirpation</td>
<td>A</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>F, 34</td>
<td>SAH, rt hemiparesis</td>
<td>rt peripheral</td>
<td>lt paraventricular</td>
<td>clipping</td>
<td>extirpation</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>F, 52</td>
<td>SAH</td>
<td>lt ICA-PCoA junction†</td>
<td>third ventricle</td>
<td>clipping</td>
<td>none</td>
<td>A</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>M, 38</td>
<td>convulsion</td>
<td>lt ICA bifurcation</td>
<td>lt parietotemporal</td>
<td>clipping</td>
<td>extirpation</td>
<td>B</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>F, 50</td>
<td>SAH</td>
<td>lt PICA ‡</td>
<td>lt cerebellar</td>
<td>resection</td>
<td>extirpation</td>
<td>A</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>F, 21</td>
<td>SAH, lt hemiparesis</td>
<td>rt ACA</td>
<td>rt frontal†</td>
<td>ligation</td>
<td>extirpation</td>
<td>A</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>M, 33</td>
<td>convulsion, SAH, lt hemiparesis</td>
<td>rt ACA</td>
<td>rt deep parietal†</td>
<td>clipping</td>
<td>extirpation</td>
<td>C</td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>M, 51</td>
<td></td>
<td>rt ACA-PCoA junction</td>
<td>rt temporal†</td>
<td>ligation</td>
<td>extirpation</td>
<td>A</td>
<td></td>
</tr>
</tbody>
</table>

†A = symptom-free or minimal symptoms; B = partially disabled but working; C = unable to work but capable of self-care; D = totally incapacitated.

Sources of hemorrhage.

*Abbreviations: AVM = arteriovenous malformation; SAH = subarachnoid hemorrhage; ACA = anterior cerebral artery; ICA = internal carotid artery; PCoA = posterior communicating artery; MCA = middle cerebral artery; PICA = posterior inferior cerebellar artery.

TABLE 2
Summary of incidence of aneurysms associated with AVM's

<table>
<thead>
<tr>
<th>Authors, Year</th>
<th>Total Series</th>
<th>Aneurysms &amp; AVM's</th>
<th>No.</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Paterson &amp; McKissock, 1956</td>
<td>110</td>
<td>3</td>
<td>2.7</td>
<td></td>
</tr>
<tr>
<td>Anderson &amp; Blackwood, 1959</td>
<td>9</td>
<td>5</td>
<td>55.6 (autopsy)</td>
<td></td>
</tr>
<tr>
<td>Cronqvist &amp; Troupp, 1966</td>
<td>150</td>
<td>13</td>
<td>8.7</td>
<td></td>
</tr>
<tr>
<td>Perret &amp; Nishioka, 1966</td>
<td>490</td>
<td>37</td>
<td>7.6</td>
<td></td>
</tr>
<tr>
<td>Suzuki &amp; O numa, 1979</td>
<td>140</td>
<td>9</td>
<td>6.4</td>
<td></td>
</tr>
</tbody>
</table>

five of nine cases (55.6%). A summary of the incidence reported in these papers is given in Table 2.

Various views have been expressed with regard to the association of aneurysms with AVM's. These theories include the following: 1) both aneurysms and AVM's are congenital vessel abnormalities; 2) due to the AVM, increased blood flow results in development of the aneurysm; and 3) their simultaneous appearance is coincidental without any causal relationship between them. In support of the second hypothesis, Shenkin, et al., recently reported a case in which an aneurysm on the main feeder to an AVM

Fig. 1. Case 1. Left: Preoperative right carotid angiogram, demonstrating an arteriovenous malformation (AVM) in the parieto-occipital region, and also an aneurysm at the junction of the pericallosal and callosomarginal artery. Right: Postoperative angiogram showing no AVM.

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reduced considerably in size due to thrombus formation after excision of the AVM. The aneurysm is not always on the main feeder of the AVM. According to the statistics of Perret and Nishioka, 43% of the aneurysms have no anatomical relationship to the AVM at all, while 37% are found on feeder arteries of the AVM, and 20% develop proximal to the origin of the main feeding artery. Nine of the 13 cases (69.2%) of Cronqvist and Troupp were found to have aneurysms on the feeders of the AVM. In our nine cases, four (44.4%) had aneurysms on the feeders, in three cases the aneurysms were sited near the origin of the AVM.
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the main feeding artery, and in two cases the aneurysms were thought to have no relationship to the AVM. Of the four patients in whom the aneurysm was located on the feeders of the AVM, three had aneurysms on the pericallosal branch of the anterior cerebral artery. Furthermore, in all three cases the anterior cerebral artery was abnormally large. Since it is known that aneurysms at this location are not common, it is thought that chronic increased blood flow may contribute to the formation of aneurysms, at least in cases of aneurysms on the feeder arteries.

The third hypothesis of etiology is supported by the frequency of aneurysm development. A high incidence of aneurysms was found by Chason and Hindman, who reported 137 aneurysm cases (4.9%) out of 2786 autopsies, and by Stehbens, who reported 77 aneurysm cases (5.6%) in 1364 autopsies. Geographical distribution and the selection of autopsy cases may influence the data, but these figures indicate that the incidence of aneurysms is high. Since from 2.7% to 8.7% of aneurysms are accompanied by AVM's, the total number of these cases is noteworthy.

Cases differ according to whether the aneurysm or the AVM brings about the first symptoms, with regard to subarachnoid hemorrhage (SAH) and convulsions. The reports of various authors indicate that symptoms of the AVM more often precede those of the aneurysm than vice versa. In cases of SAH, there are surgical problems concerned with the origin of hemorrhage, and it is generally difficult to make the determination based solely upon symptoms. According to Perret and Nishioka, out of 29 cases of SAH, the hemorrhage originated slightly more frequently from the AVM (nine cases); in seven cases the hemorrhage originated at the aneurysm, and in 13 cases its origin was unclear. Of our nine cases, seven were found to have SAH. From the findings at surgery, symptoms, location of the lesion, and the nature of its development, the hemorrhage was thought to have originated in the AVM in three cases, from the aneurysm in three cases, and from both in one case (Case 2). This last patient had hemorrhage and intracranial murmur at the time of onset of symptoms, and finally developed oculomotor paresis. It appears to have been an interesting case in which hemorrhage had occurred before surgery from both the aneurysm and the AVM.

It is undoubtedly best to perform radical surgery on both the aneurysm and the AVM, but reports to this effect are not numerous. Of 13 cases reported by Cronqvist and Troupp, only four received radical surgical treatment; of these, only two patients were treated for both the aneurysm and the AVM. In the other two patients either the aneurysm or the AVM was treated; one of these patients subsequently died from hemorrhaging of the untreated lesion. Of the 29 hemorrhage cases reported by Perret and Nishioka, 14 were treated surgically: the AVM alone was treated in seven cases, the aneurysm alone in two cases, and both in five cases. A high mortality rate was found among the cases treated conservatively, namely, nine deaths in 15 cases (60%).

In our nine cases, all but one case with an AVM near the third ventricle were treated by radical surgery, producing favorable results with no mortality. Surgery should be directed to treatment of the source of hemorrhage, but determination of the source preoperatively is often difficult and as a matter of principle we have been treating both lesions in a single operation. The administration of 500 to 1000 cc of 20% mannitol just before craniotomy makes it possible to extend the period of temporary occlusion of the parent artery of an aneurysm and main feeders of an AVM to 40 to 50 minutes. This temporary occlusion minimizes the danger of hemorrhage and simplifies the surgical procedure.

Since the number of cases with this lesion is still small and reports of its natural history are few, questions remain concerning its treatment. Insofar as 1) there are two possible sources of hemorrhage, 2) the mortality rate following conservative treatment is 60%, and 3) even when surgery is performed on either the aneurysm or the AVM, there is danger of death from hemorrhage of the untreated lesion, it is desirable to treat both lesions in a single radical surgical operation.

Summary

1. From 1959 to 1975 we have treated 140 cases of AVM, nine of which (6.4%) had associated cerebral aneurysms.
2. Among these nine cases, all but a small, deep-seated AVM were cured by means of radical surgery on the aneurysm and total extirpation of the AVM.
3. It is thought that effective treatment is possible by performing surgery on both the aneurysm and the AVM, with temporary clipping of the feeders of both, thereby reducing the danger of hemorrhage.
4. The administration by infusion of 500 to 1000 cc of 20% mannitol solution just before craniotomy is recommended to prolong the occlusion time of intracranial arteries under normotensive and normothermic anesthesia.

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