The association of intracranial aneurysms and arteriovenous malformation of the brain

Case report

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A case of intracranial multiple aneurysms associated with an arteriovenous malformation (AVM) is described. Three aneurysms were found arising from an enlarged anterior cerebral artery feeding an AVM. In spite of the fact that two of these aneurysms received no surgical treatment, they disappeared almost completely several months after excision of the AVM. Seventy-three previously reported cases of cerebral aneurysms associated with AVM's are reviewed, and the effect of hemodynamic stresses on the development of these aneurysms is summarized.

KEY WORDS • intracranial aneurysm • arteriovenous malformation • hemodynamics

The association of intracranial aneurysms and cerebral arteriovenous malformations (AVM's) has frequently been reported, and three hypotheses that may explain the mechanics of this association have been proposed. One is that the increased hemodynamic stresses resulting from the AVM's may play a decisive part in aneurysm formation. This hypothesis is strongly supported by the findings in our case as well as the 73 cases we have reviewed in which the effect of hemodynamic stresses on aneurysm formation was studied.

Case Report

This 39-year-old man appeared to be perfectly well until he was found unconscious in bed. He was immediately taken to a local hospital. On admission, he was semicomatose with Cheyne-Stokes respiration and fixed pupils. One hour later he became stuporous. The left pupil was larger than the right, and both reacted to light only slowly. There were bilateral extensor plantar reflexes. Several days after admission, he regained consciousness but had right hemiplegia and aphasia. Blood pressure was 150/80 mm Hg. Lumbar puncture revealed bloody cerebrospinal fluid. Left carotid angiography disclosed an AVM in the left frontal lobe fed by the internal frontal arteries, which were enlarged. Three aneurysms arising from the enlarged feeding arteries were also disclosed; one was located at the junction of the callosomarginal and internal frontal arteries, another at the junction of the callosomarginal and frontopolar arteries, and the last at the junction of the pericallosal and callosomarginal arteries (Figs. 1A and 2A). Marked displacement of the distal anterior cerebral artery and the lack of vasoospasm around the aneurysms suggested an intracerebral hematoma, probably due to bleeding from the AVM.

The patient was transferred to our clinic 6 days after the onset of symptoms. Through a left craniotomy, the AVM was totally extirpated and a hematoma, 30 ml in volume, was evacuated. The most peripherally situated aneurysm on the callosomarginal artery was also successfully clipped. The other two aneurysms were not treated surgically because they did not have distinct necks.

The postoperative course was uneventful, although right hemiplegia and aphasia persisted for a few months. A second carotid angiogram taken 12 days after surgery revealed that both the AVM and the most peripherally situated aneurysm were completely abolished, while the unoperated aneurysms remained
unchanged in size and shape (Figs. 1B and 2B). A third carotid angiogram taken on the 74th postoperative day showed reduction in the size of the unoperated aneurysms and a decrease in the caliber of the previously enlarged feeding arteries (Figs. 1C and 2C). The patient was discharged with minimal hemiparesis and returned to his job as a laborer. On the fourth carotid angiogram taken 6 months after surgery, the aneurysms had almost disappeared and the arteries were normal in size (Figs. 1D and 2D). The serial change of aneurysms traced from these angiograms is schematically represented in Fig. 2. It was very interesting that the diminished aneurysms ran smoothly into the arteries (Figs. 1D and 2D).

Analysis of Reported Cases

Seventy-three previous cases of intracranial aneurysms associated with AVM’s
were reviewed in order to assess the effect of hemodynamic stresses on aneurysm formation. Cases with incomplete hemodynamic descriptions were excluded. The 112 aneurysms detected in 74 cases, including ours, were divided into the following three groups depending on the site of the aneurysms and the blood supply to the AVM’s:

Group A: Aneurysms located on a major feeding artery itself
Group B: Aneurysms located on the proximal portion of the feeding system, such as the ipsilateral internal carotid artery, the anterior communicating artery, and the contralateral internal carotid artery that were feeding the AVM’s
Group C: Aneurysms located on arteries hemodynamically unrelated to AVM’s.

Abnormally enlarged arteries verified only at autopsy without angiography were also interpreted as feeding arteries or feeding systems, and the aneurysms on these arteries were classified under Group A or B.

The cases are summarized in Fig. 3. The patients included 44 males, and 28 females; in two cases the sex was not mentioned. Their ages ranged from 1 to 74 years, with an average of 42.5 years. The average age of patients with aneurysms alone was higher than that of patients with combined anomalies, and that of patients with AVM’s alone was lower. Twenty-one patients (28%) had multiple aneurysms. The location of aneurysms is shown in Fig. 4. There were differences in the location of the aneurysms between the cases associated with AVM and those not so associated. Eighteen aneurysms were present on the distal anterior cerebral artery, 15 of which were on the feeding artery itself. The incidence of distal anterior cerebral artery aneurysms in this series (16%) was much higher than that in the series of aneurysms alone (5% in all Japan in 1969).28

On the other hand, nine aneurysms existed on the peripheral portions of the lenticulostriate, anterior choroidal, posterior choroidal, superior cerebellar, anterior inferior cerebellar, and posterior inferior cerebellar arteries, where aneurysms did not usually develop. Eight of them were present on the feeding artery itself. There were 45 aneurysms in Group A, 41 in Group B, and 26 in Group C. Eighty-six of the 112 aneurysms (77%) were on arteries hemodynamically related to AVM’s. From these results it may be surmised that hemodynamic stress was the factor most likely to be responsible for the formation and/or the development of aneurysms in these cases.

Discussion

Intracranial aneurysms are often associated with AVM’s.1-14,16-27,29-31 It has been discussed whether there is any special relationship between the two

FIG. 1. Left carotid angiograms before (A), and 12 days (B), 74 days (C), and 207 days (D) after surgery. The unoperated aneurysms gradually reduced in size and the arteries became almost normal.
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Fig. 2. The serial change of aneurysms traced from the arteriograms in Fig. 1. Note the almost complete disappearance of the two unoperated aneurysms and the remarkably smooth contour of these aneurysms with the arteries.

Fig. 3. Summary of 112 aneurysms in 74 cases.
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FIG. 4. Location of 112 aneurysms in 74 cases with associated arteriovenous malformations (AVM's). Each circled numeral shows the number of aneurysms hemodynamically related (white) and unrelated (black) to the AVM: 77% of aneurysms were located on the arteries hemodynamically related to the AVM. The incidence of aneurysms on the distal part of the arteries was much higher than in cases without AVM's.

different kinds of lesions or not, and three hypotheses have been propounded in an attempt to explain the nature of this association. The three hypotheses are as follows: 1) both lesions are congenital in origin, and the association may be an expression of multiple disorders of vascular development; 2) the association is a coincidence; and 3) aneurysms are caused by hemodynamic stresses resulting from the presence of an AVM.

In a few cases, only one explanation was given. It is true that each of these hypotheses is based on bits of evidence. However, the first hypothesis cannot be accepted because intracranial aneurysms are not purely congenital in origin. A medial defect of the arterial wall may be congenital and it can be an important factor in aneurysm formation, but it is found in many parts of the arterial bifurcation where aneurysms do not develop. Another objection is raised against this hypothesis on the ground that aneurysms rarely develop in childhood. The second hypothesis is incomplete, although it is accepted in some of the cases. The incidence of aneurysms occurring in association with AVM's is 6% to 9%, according to the recent clinical studies covering a large number of AVM's and it is higher than that of aneurysms alone (1% to 5%) in a general population. This fact definitely suggests that there may be a special relationship between the two different sorts of lesions.

The role of hemodynamic stresses in the pathogenesis of aneurysms has been mentioned by many authors. For example, Hashimoto, et al., stated emphatically that cerebral aneurysms were produced only when hemodynamic stresses were increased, as by experimental carotid ligation. Likewise, the present analysis of the reported cases revealed a close relationship between the site of the aneurysms and the supply of blood to the AVM's. In view of the fact that a comparatively high incidence of aneurysms was found on the distal anterior cerebral artery and the peripheral parts of the anterior and posterior choroidal, the lenticulostriate, the superior cerebellar, and the anterior and posterior inferior cerebellar arteries, it can be postulated that hemodynamic stresses may play a vital part in aneurysm formation. Thus, the third hypothesis is considered to be the most plausible.

Our observation that two unoperated aneurysms disappeared after excision of the AVM supports the third hypothesis. Shenkin, et al., reported a patient whose internal carotid artery aneurysm decreased remarkably in size after removal of the AVM fed by this artery. Takara, et al., described a case in which an aneurysm of the left anterior inferior cerebellar artery coexisted with the AVM fed by the same artery. In their case, both the aneurysm and AVM disappeared spontaneously 4 months after nonradical surgery, that is, wrapping of the aneurysm without surgical treatment of the AVM. They inferred that the disappearance of the aneurysm might be due to the decreased blood flow of the parent artery secondary to the preceding regression of the AVM. Although no more than two of the cases we reviewed were similar to ours, it can be speculated that aneurysms situated on arteries feeding AVM's may possibly be cured merely by means of excising the AVM's concerned.

Spontaneous regression of aneurysms, which has rarely been reported, is thought to be brought about by thrombosis of the aneurysms. In our case, the unoperated aneurysms almost completely disappeared, and the configuration of the diminished aneurysms ran smoothly into the feeding arteries. It is unlikely that intraluminal thrombosis occurs so extensively as to involve feeding arteries and so smoothly as observed in our case. It is thought, therefore, that regression may most probably be attributable to the contraction and/or intimal thickening of arteries. The possibility is thus raised that some aneurysms may be cured by reducing the blood flow of a parent artery only. It may be argued that carotid ligation does not usually give rise to a reduction in the size of an internal carotid artery aneurysm, but this is well explained by the fact that carotid ligation does not always cause a reduction of the blood flow of the artery. By contrast, excision of an AVM surely leads
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to a reduction of the blood flow in the artery feeding it, so that aneurysms on such a vessel can heal without surgical treatment.

References
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