CEREBRAL ANEURYSMS WITH OTHER INTRACRANIAL PATHOLOGY

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Subdural and intracerebral hematomas are recognized surgical lesions which may accompany a rupture of cerebral aneurysm. The incidence of multiple cerebral aneurysms has been reported between 9 and 21 per cent in large series of cases of this disease. Congenital anomalies elsewhere in the body are known to occur with cerebral aneurysm, such as coarctation of the aorta and polycystic kidney. In a study of 250 consecutive adult patients in our series who had cerebral angiography, cerebral aneurysms were found in 3 patients who had independent cerebral lesions which included an arteriovenous aneurysm, cerebral cyst and a cerebral hematoma. The diagnostic problems associated with these latter combinations are illustrated.

CASE REPORTS

Case 1. A 50-year-old white male was treated in another hospital 4 months prior to admission for arterial insufficiency with ischemic neuropathy of both lower limbs. Aortography, lumbar sympathectomy, aortic thrombectomy, and four attempts at vascular grafting of aorta, left external iliac or left common femoral arteries were performed. Operations were complicated by infection and rupture of grafts and the development of gangrene of the lower extremities requiring amputation at the thigh on the left leg and below the knee on the right leg.

Physical findings were normal except for the amputated lower extremities and a granulating ulcer on the stump of the left thigh. Two weeks after application of a split-thickness skin graft to the stump of the left thigh, there was elevation of the patient’s temperature with physical signs of congestion of the right lung. He was afebrile after treatment for 4 days with tetracycline, 250 mg. 4 times a day, when he had a grand mal seizure, became comatose and had a left hemiplegia. Lumbar puncture revealed grossly bloody spinal fluid.

Right carotid angiogram revealed a 3 mm. aneurysm on a branch of the right callosal marginal artery (Fig. 1). Elevation of the right middle cerebral artery was not recognized in the initial study of the angiogram.

Twelve hours after the onset of coma, the aneurysm, which measured 1×0.8 cm., on the right callosal marginal artery was excised by right frontal craniotomy. Although subarachnoid hemorrhage was present in the operative area, signs of rupture of this aneurysm were absent. Internal decompression of the tip of the right frontal lobe was performed to alleviate edema of the brain.

† Angiograms were made in all cases by the percutaneous technique using injections of 10 cc. of 50 per cent Hypaque Sodium.
The patient expired 3 days after surgery. Pathological diagnosis of the surgical specimen was berry aneurysm of cerebral artery.

Necropsy disclosed subarachnoid hemorrhage over the cerebral hemisphere and base of the brain. A surgical defect, which measured 3 cm. in diameter, was present in the right frontal lobe. An independent intracerebral hematoma, which measured 5 cm. in greatest diameter, extended from the right Sylvian fissure into the anterior portion of the putamen and caudally into the posterior parietal lobe. Microscopic study of this hematoma revealed areas which sometimes contained small central masses of fibrin and a few segmented neutrophils. One artery from the clot had subendothelial infiltration with segmented neutrophils. All three leaflets on the ventricular surface of the aortic valves contained red-brown, large clot. Microscopic sections of the aortic valve revealed fibroblastic proliferation of the valve leaflets, large attached clot which showed early organization, and multiple clumps of bacteria.

Principal pathological diagnoses were: acute bacterial endocarditis of aortic valve; acute arteritis of cerebral vessel; subarachnoid and intracerebral hemorrhage; berry aneurysm (surgical specimen) of cerebral artery; cerebral edema; arterial homografts of abdominal aorta, iliac arteries, and right femoral artery.

Case 2. A 33-year-old right-handed male had been hospitalized on several occasions elsewhere during the year prior to admission for severe headache, loss of consciousness, and intermittent blindness in the left field of vision. He was admitted after a transient period of unconsciousness.

On admission neurological findings were within normal limits except for nuchal rigidity. Lumbar puncture revealed grossly bloody spinal fluid. Right carotid angiogram demonstrated nonfilling of the right anterior cerebral artery and visualized a right occipitoparietal arteriovenous aneurysm filled from the middle cerebral
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Fig. 2. Case 2. Right carotid angiogram demonstrating right occipitoparietal arteriovenous aneurysm (arrow) which is filled by right middle cerebral artery.

artery (Fig. 2). Left carotid angiogram disclosed bilateral filling of the distal anterior cerebral arteries with demonstration of the above described arteriovenous aneurysm filled from the distal right anterior cerebral artery. Aneurysm was suspected in the proximal region of the anterior cerebral artery on the left side (Fig. 3),

Fig. 3. Case 2. Left carotid angiogram demonstrating suspected aneurysm (arrow) in proximal region of anterior cerebral artery.
FIG. 4. Case 2. Coronal section of brain demonstrating aneurysm of right anterior cerebral artery (top arrow) and intraventricular hemorrhage. Coronal section of pons shows arteriovenous aneurysm in its base (lower arrow).

but this could not be verified by repeated angiography 2 weeks later. Nine days after this examination he had a second subarachnoid hemorrhage, following which he had bilateral severe retinal hemorrhages, left homonymous hemianopsia, left facial weakness, and sensory loss in the left arm and leg.

Operation was performed under hypothermia 3 days after the second subarach-
noid hemorrhage. The arteriovenous aneurysm was apparent on the lateral surface of the posterior parietal area. Branches of anterior, middle and posterior cerebral arteries were transected in the block resection of the arteriovenous malformation and right parietal lobe of the brain down to the falx and tentorium. The major venous channels were found to communicate with the vein of Galen. This vascular lesion was a purplish-red lobulated mass, which measured 6 X 6 X 2 cm. Areas of old hemorrhage were encountered in the resection.

For 48 hours following surgery he was alert and talking, when he suddenly become comatose and expired.

Necropsy revealed an aneurysm, which measured 1.0 cm. in diameter, on the right anterior cerebral artery. The aneurysm was filled with blood clot. Hemorrhage extended from this lesion into the right frontal lobe and adjacent lateral ventricle (Fig. 4). A small arteriovenous aneurysm was present in the base of the pons. The right occipitoparietal lesion had been excised and no bleeding point could be found in the operative area.

Case 3. A 35-year-old male had a 1-year history of bifrontal headaches and increasing difficulty in reading.

Examination revealed a right homonymous hemianopsia, early papilledema, and impairment of position sensation in the right foot.

Ventriculography, attempted through a left posterior parietal burr hole, outlined the presence of a left parieto-occipital cyst measuring 8 X 4 X 5 cm. Right carotid injection with contrast medium revealed an aneurysm of the supraclinoid portion of the right internal carotid artery (Fig. 5).

Left occipital osteoplastic craniotomy was performed and subarachnoid drainage of multiple ependymal-lined cysts in the parieto-occipital area was accomplished by cortical incision.

The patient’s postoperative course was uneventful and visual fields were improved 3 months after operation.

DISCUSSION

Table 1 is a summary of the reported cases of combinations of cerebral aneurysms with arteriovenous aneurysm and brain tumor which we have encountered in the literature. The pathology of intracranial arteriovenous aneurysm has occasioned considerable dispute in the past and as a consequence the nomenclature is somewhat confused. However, synonyms in vogue include “intracranial angioma,” “arteriovenous malformation” and “angiomatous malformation.” Three patients in the series of 110 cases of intracranial angioma reported by Paterson and McKissock28 had “true” intracranial arterial aneurysms in addition to their angioma. They used the adjective “true” to distinguish them from pseudoaneurysmal formation which may develop from an angioma. Gould et al.13 indicated that they had encountered 3 patients with the combination of aneurysm and arteriovenous aneurysm, but they did not describe them in their series of 41 cases of arteriovenous malformations. On the other hand, this combination was not mentioned in studies of series of arteriovenous aneurysms as reported by numerous authors.1,4,6,10—12,14,15,17,19—25,29—34,36—39

Heppner and Lechner16 have emphasized the diagnostic problems in
simultaneous occurrence in the brain of aneurysm and tumor. They reported the case of a patient who had a combination of supraclinoid aneurysm of the left internal carotid artery with glioblastoma of the left temporal and occipital lobes. *In vivo* only the aneurysm had been diagnosed and this was considered the cause of the neurological symptoms. A full explanation was possible only at autopsy. This was similar to the diagnostic difficulties in our Case 1, in which neurological findings and subarachnoid hemorrhage suggested that rupture of the demonstrated aneurysm had occurred. Failure to recognize elevation of the middle cerebral artery in the angiogram precluded diagnosis of the existing independent intracerebral hematoma in the posterior parietal and temporal areas. In Case 2, the presence of clot in the aneurysm itself probably accounted for the difficulty in visualization of the suspected aneurysm by repeated angiography. While the clinical signs and symptoms had suggested hemorrhage from the arteriovenous aneurysm, the therapeutic failure and demise were related to the rupture of the cerebral aneurysm. In Case 3 the presence of an asymptomatic cerebral aneurysm was disclosed in the angiographic study and subsequent clinical course. Therapy of the cerebral cyst, however, could have been complicated by rupture of the aneurysm when the intracranial pressure was reduced.

**Table 1**

*Reported cases of arteriovenous aneurysm and brain tumor with an associated cerebral aneurysm*

<table>
<thead>
<tr>
<th>Author</th>
<th>Site of Aneurysm</th>
<th>Independent Intracerebral Lesion</th>
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<tbody>
<tr>
<td>Arieti</td>
<td>Left middle cerebral artery</td>
<td>Multiple meningioma</td>
</tr>
<tr>
<td>Arieti &amp; Gray</td>
<td>Supraclinoid—left internal carotid artery</td>
<td>Left parietal angiomata</td>
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<tr>
<td>Budin &amp; Obrda</td>
<td>Left internal carotid artery</td>
<td>Left acoustic neurinoma</td>
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<tr>
<td>Caram</td>
<td>Anterior communicating artery</td>
<td>Arteriovenous malformation of left anterior temporal and inferior frontal areas</td>
</tr>
<tr>
<td>Descun et al.</td>
<td>Callosal marginal branch of left anterior cerebral artery</td>
<td>Arteriovenous aneurysm on pericallosal branch of left anterior cerebral artery</td>
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<tr>
<td>Heppner &amp; Lechner</td>
<td>Supraclinoid—left internal carotid artery</td>
<td>Glioblastoma of left temporal and occipital lobes</td>
</tr>
<tr>
<td>Lau</td>
<td>Multiple—right internal carotid artery; gross anomaly circle of Willis</td>
<td>Arteriovenous aneurysm of left parieto-occipital lobe</td>
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<tr>
<td>Paillas et al.</td>
<td>Left middle cerebral artery</td>
<td>Arteriovenous aneurysm of left frontoparietal area</td>
</tr>
<tr>
<td>Paterson &amp; McKissock</td>
<td>Multiple—both on anterior and middle cerebral arteries on left</td>
<td>Angioma of left Sylvian fissure</td>
</tr>
<tr>
<td>Paterson &amp; McKissock</td>
<td>Multiple—two on right internal carotid artery</td>
<td>Right parietal angioma</td>
</tr>
<tr>
<td>Paterson &amp; McKissock</td>
<td>Right posterior inferior cerebellar artery</td>
<td>Angioma of left cerebellar hemisphere</td>
</tr>
<tr>
<td>Vieten</td>
<td>Left middle cerebral artery</td>
<td>Arteriovenous malformation of left occipital area</td>
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The congenital origin of arteriovenous aneurysm and cerebral berry aneurysm has long been suspected. The rarely encountered combination of arterial aneurysm and arteriovenous aneurysm may be related to a difference in their possible embryological origin. Embryological and clinical studies have suggested that the arterial aneurysm commences in errors in fusion and absorption in the normal dynamics of the developing arterial system. This phenomenon occurring in several loci in the developing arterial system could account for the greater frequency of multiple cerebral aneurysms than the combination of cerebral aneurysm with arteriovenous aneurysm. The embryological studies by Padget are revealing for the possible difference in the origin of the arteriovenous aneurysm as noted in our Case 2. These studies demonstrate an intimate relationship in the endothelial stages of the posterior choroidal artery with its accompanying vein; fistulous formation in this stage of development with reversal or disturbance of flow of blood in adjacent vessels in embryonic life and spread during the subsequent expansion of the cerebral hemisphere could account for the involvement of anterior, middle and posterior cerebral arteries as seen in the adult lesion.

SUMMARY

Three patients are reported who had independent intracranial lesions in addition to a cerebral aneurysm. These included a case of arteriovenous aneurysm, cerebral cyst and independent intracerebral hematoma. Therapeutic failures resulted in these cases when these combinations were not recognized.

REFERENCES