

## Coexistence of Bilateral Aneurysms of the Internal Carotid Arteries and an Arteriovenous Malformation of the Left Frontal Lobe

Román Garza-Mercado, M.D., Ricardo A. Rangel, M.D., and Juan F. Garza-Vázquez, MD

Divisions of Neurosurgery, and Neurology, Facultad de Medicina y Hospital Universitario "J. Eleuterio González," Universidad Autónoma de Nuevo León, Monterrey, Nuevo León, México

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A 44-year-old right-handed man was hospitalized because of a subarachnoid hemorrhage. He was drowsy on admission and exhibited a left partial third nerve palsy, right hemiparesis, and selective motor aphasia. A large subcortical arteriovenous malformation involving the left frontal lobe was demonstrated by angiography. The malformation filled from either carotid system and from the vertebrobasilar system as well. In addition, there were two bilaterally symmetrical (mirror) aneurysms of each internal carotid artery. Computed axial tomography revealed a left intratemporal-intraventricular hematoma. Deterioration of the patient's clinical condition occurred a few days after admission. Improvement was obtained after removal of the hematoma at a left subtemporal craniectomy, at which time the ruptured aneurysm arising from the left internal carotid artery was clipped. Sixteen months later, the intact aneurysm of the right internal carotid artery was clipped, and the arteriovenous malformation was radically excised through a bifrontal (Pool) craniotomy with microsurgical technique and with the aid of intraoperative angiography. The patient remained with a fixed neurological deficit, as observed preoperatively.

**KEY WORDS:** Aneurysm, Arteriovenous malformation; Intracerebral hematoma; Intraoperative angiography; Multiple aneurysms; Subarachnoid hemorrhage

The simultaneous association of intracranial aneurysms and arteriovenous malformation is a fairly rare clinical condition. In 1959, Boyd-Wilson [7] analyzed 17 such cases from the literature, three of his own included, and

in 1981, Hayashi et al [18] accounted for 74. Such coexistence is even rarer when the arteriovenous malformation occurs with symmetrically opposite aneurysms of internal carotid arteries in a mirrorlike fashion, as in the case to be presented. Credit is given in the literature [7] to Walsh and King [40] as the first to record in 1942 a case of coexisting aneurysm and arteriovenous malformation. Their patient, a 50-year-old black man, was hospitalized because of subarachnoid hemorrhage. He had a 4-year history of intermittent right frontal headache, right third cranial nerve palsy, and right homonymous hemianopsia. The clinical diagnosis of a right supraclinoid aneurysm was corroborated at the operation, but the aneurysm was found thrombosed. The patient died a few days later, and at necropsy a ruptured arteriovenous malformation of the left occipital lobe was disclosed, with intraventricular and subarachnoid hemorrhages. The aneurysm of the right internal carotid artery was unruptured and filled with a thrombus, and a symmetrical, although smaller, aneurysm was also found on the opposite internal carotid artery. Walsh [39] describes this patient as case 377 in his *Clinical Neuro-Ophthalmology*. The incidence of coexisting aneurysm and arteriovenous malformation has been recorded from 2.7% [31] and 3% [30] to 9.3% [19]. In 490 patients with arteriovenous malformation, Perret and Nishioka [32] found 37 cases of coexisting aneurysm (7.55%). Of the 34 of their patients studied in detail, there were six with two aneurysms, two with three, one with six, and another one with "multiple small aneurysms." In reviewing a series of 112 aneurysms found in 74 patients, Hayashi et al [18] noted 28% to have multiple aneurysms. In reporting a personal series of 30 large (namely, larger than 2 cm [30]) arteriovenous malformations, 20 of which were submitted to microsurgical treatment, one of the authors (R.G.-M.) found two patients with associated intracranial aneurysms (6.6%) [13]. One of these two patients had one aneurysm, the case hereby presented had two.

Address reprint requests to: Román Garza-Mercado, M.D., Aleutianas 396, Col. V. Hermosa, Monterrey, N.L., México.

## Case Report

A 44-year-old right-handed man was admitted to the Hospital Universitario on July 8, 1978. Two hours earlier he had been awakened from his sleep in the middle of the night by an intense headache of sudden onset. Although he could get up, only mumbling of unintelligible expressions were heard by the relatives, and a right-sided weakness was obvious. He lapsed into a short period of unconsciousness before his admission to the hospital. The past history was significant only in that partial seizures involving the right side had occurred intermittently since age 11 years, when a diagnosis of meningitis (subarachnoid hemorrhage?) had been made. There was no history of hypertension, renal dysfunction, or tobacco consumption.

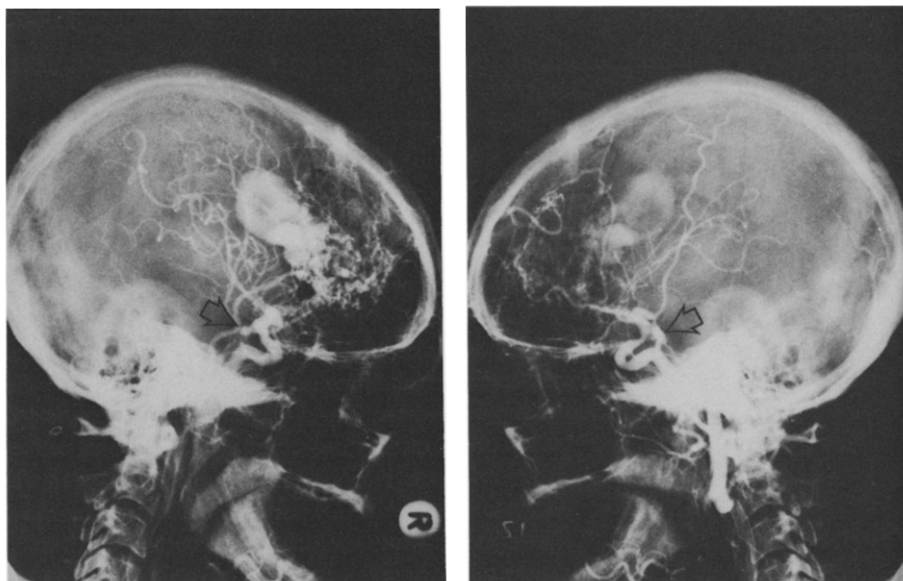
On examination, the patient was drowsy. He responded to mild painful stimuli by flexion of the left extremities. His blood pressure was 140/70 mmHg, pulse 74 beats per minute, temperature 36.5°C, respiratory rate 17 breaths per minute. He was unable to speak. There was a paresis of the lower right side of his face, and a partial left third nerve palsy. The left pupil was slightly dilated but reacted to light. The optic fundi were normal. There was a spastic hemiparesis of the right side, with increased deep-tendon reflexes and a right Babinski sign. Moderate stiffness of the neck was noticed. No lumbar puncture was performed then. Admitted to the neurology division, the patient improved by the next day, and he was well enough to attend simple commands. It was now evident that he had a motor aphasia. Laboratory data and x-ray studies of the skull and chest were normal. Angiograms of the bilateral carotid and right vertebral arteries (via a femoral artery) were made under local analgesia on the fifth day after admission. A large subcortical arteriovenous malformation containing a bilobulated varicose dilatation was seen to occupy most of the left frontal lobe. It was fed predominantly from the left anterior cerebral artery, with a few branches from the left middle cerebral artery. However, the malformation opacified equally well when the right carotid artery was injected, through a dilated anterior communicating artery, and it received an important contribution from the vertebrobasilar system also via branches of the left posterior cerebral artery. After two seconds, the malformation drained into both the superior longitudinal sinus and the deep venous system. The left middle cerebral artery was displaced upwards and medially. In addition, bilaterally symmetrical opposite aneurysms were observed on each internal carotid artery, at the level of the posterior communicating artery (Figure 1). Computed axial tomography performed on the seventh day of admission revealed an intratemporal-intraventricular hematoma on the left side (Figure

1). A neurosurgery consultation was then requested because of worsening of the patient's clinical condition. The patient was drowsier than before the computed tomography scan, and the third nerve palsy was now complete. The patient was promptly taken to the operating room.

On the evening of July 15, 1978, an emergency left subtemporal craniectomy was performed under general endotracheal anesthesia. The dura mater was tense and not pulsating, and it was blue in color. After aspiration of 60 mL of old liquified blood through a ventricular needle, pulsation of the dura mater was observed. A large intratemporal clot was removed through a 3-cm horizontal cortical incision in the second temporal gyrus after incising the dura mater. The hematoma, composed of dark-colored clots of different densities, was easily aspirated. The hematoma had broken into the ipsilateral temporal ventricular horn, and extended into the mesial portion of the temporal lobe. Although some abnormal vessels from the malformation were visualized, it seemed that the clot did not extend into the frontal lobe. Gentle retraction of the now-collapsed brain allowed exposure of the aneurysm arising from the left internal carotid artery, and occlusion of its neck with a curved Mayfield clip. It was obvious that the bleeding had occurred from rupture of the aneurysm and not from the arteriovenous malformation. It was felt unwise to attack the malformation at this time, so the wound was closed, leaving the dura mater open. The patient regained consciousness by the next day. He improved enough to understand simple commands, but a selective motor aphasia persisted. The hemiparesis of the right side and the complete left third nerve palsy remained unabated when he was discharged from the hospital on September 3, 1978, after postoperative angiography of the bilateral carotid and right vertebral arteries. The aneurysm on the left internal carotid artery was excluded from the circulation, but the malformation, and the aneurysm on the right internal carotid artery had the same characteristics as before.

The patient was hospitalized again from November 28, 1978, through January 24, 1979, for treatment of recurrent bouts of pyelonephritis. The neurological signs remained fixed. Gelfoam embolization of the malformation via the femoral artery was unsuccessful.

The patient was admitted to the hospital for the third time on October 25, 1979, for further neurosurgical treatment. There had been no recurrent bleeding episodes, the seizures had been under therapeutic control, and the neurological deficiency remained stationary. The patient was cheerful, and understood simple commands in spite of his selective aphasia. The laboratory data were normal. Repeat angiography of the bilateral carotid and right vertebral arteries showed both the arteriovenous



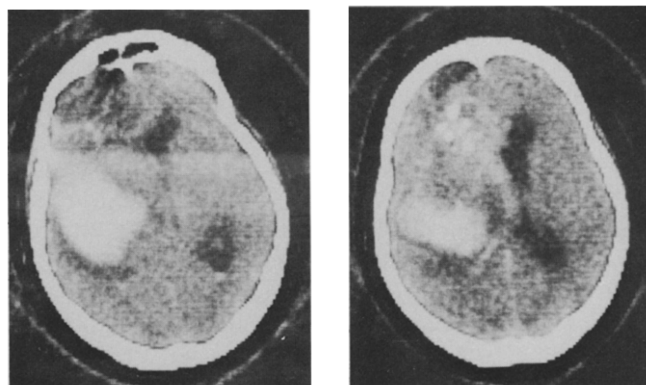
**Figure 1.** Bilateral carotid arteriograms, lateral views. The arteriovenous malformation of the left frontal lobe is seen to have opacified equally well from the right (left) and the left (right) carotid systems. The bilaterally opposite saccular aneurysms of the internal carotid arteries are clearly shown (arrows).

malformation and the aneurysm on the right internal carotid artery unchanged.

On November 13, 1979, 16 months from the initial treatment, a bifrontal (Pool) craniotomy was performed under general endotracheal anesthesia. Exposure of the optic nerve and the internal carotid artery was accomplished bilaterally after ligation and section of both the superior and the inferior longitudinal sinuses, and retraction of the frontal lobes. Under microscopy the neck of the intact aneurysm of the right internal carotid artery was occluded with a straight Mayfield clip. The anterior communicating artery and the left anterior cerebral artery were grossly enlarged. They had an estimated three-fold increase in diameter as compared with the right anterior cerebral artery. Temporary occlusion of the left pericallosal artery with a Mayfield clip did not seem to significantly reduce the vascularity of the malformation, as seen in a single intraoperative angiogram of the left carotid artery. The malformation was then radically excised. A repeat intraoperative single angiogram of the left carotid artery showed absence of the malformation, with preservation of the parent vessels, as displayed also on the postoperative cerebral panangiography. The patient was discharged on December 1, 1979 with no increase in his preoperative neurological deficit. However, he died at home on June 21, 1980, of bronchopneumonia.

### Discussion

The coexistence of intracranial aneurysms and arteriovenous malformations has been reported by several authors [1,3,4,7-9,11,13,14,18,19,22,23,31,37]. The three basic theories are reviewed by some of them [7,



**Figure 2.** Computed axial tomography. An intratemporal-intraventricular hematoma is observed on the left side. Postcontrast (Conray) film (right) shows filling of abnormal vessels of the malformation.

11,18,19,23,31,37]. Boyd-Wilson [7] considered their simultaneous association as mere coincidental lesions in the same patient, but the somewhat higher incidence of aneurysms coexisting with arteriovenous malformations (2.7 to 9.3%) than in the general population (1 to 5.6%) [2,10,18,26] has been interpreted as suggestive evidence of multiple congenital abnormalities [4,18]. Both aneurysms [2] and arteriovenous malformations [21] may develop from congenital anomalies. Most authors, however, side with the hydrodynamic stress theory, as initially expressed by Paterson and McKissock [31]. The role of altered hemodynamics in the pathogenesis of intracranial arterial aneurysms finds some support in the experimental work of Hassler [16] in rabbits, and of Hashimoto et al [15] in hypertensive rats. Induction of

new aneurysmal bulging was observed when one carotid artery was ligated in the animal's neck, and in the basilar system when the carotids were ligated on both sides. Clinical evidence of hydrodynamic influence is suggested also by the reduction in size or disappearance of associated intracranial aneurysms after removal of an arteriovenous malformation alone [18,23,36], albeit the experience of others has been entirely opposite, that is, aneurysmal rupture has occurred after resection of an associated arteriovenous malformation [22,23]. Furthermore, in a large proportion of cases the aneurysm has been located in hemodynamically related arteries, either on the main arterial feeder or in the proximal afferent arterial system. Hayashi et al [18], and Suzuki and Onuma [37] found them so related in 77% of cases, although Perret and Nishioka [32] reported them to be so located in only 53% of cases.

Multiple aneurysms represent about 20% of all intracranial aneurysms [25], but figures as high as 31.4% have been recorded [10]. The strong tendency for multiple aneurysms to occur oppositely and symmetrically, particularly when the internal carotid arteries or the middle cerebral arteries are so affected, has been recognized by several authors [2,6,10,16,27,28]. Locksley [25] has noticed that 47% of all double aneurysms were located contralaterally to each other. When one aneurysm arises on an internal carotid artery, the opposite aneurysm will be on the internal carotid artery in 49% of cases, with a better than 3:1 chance for being symmetrically disposed. Our patient had symmetrical opposite internal carotid artery aneurysms, the left one being symptomatic.

We feel that both aneurysms and arteriovenous malformations, whether independent or associated, should be treated surgically. The rationale and safety for operating on asymptomatic and incidental aneurysms has been reemphasized by Salazar [34] and Wirth et al [41] following the lead of others [12,20,27,28,35]. In our patient, the malformation was located in the left (dominant) frontal lobe, but it had strong vascular afferents from both internal carotid artery systems. The opposite aneurysms arose from vessels hemodynamically related to the malformation. The rupture of the aneurysm on the left internal carotid artery was responsible for the subarachnoid hemorrhage and the intratemporal-intraventricular hematoma. Neither the malformation, nor the aneurysm on the right internal carotid artery had ruptured at the time of the operations.

The usefulness of intraoperative cerebral angiography in the treatment of arteriovenous malformations and aneurysms has been well recognized [5,24,29,38]. A radical removal of the malformation can be documented at the time of the operation, as shown in our patient.

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