

# "Giant Pneumoaneurysm" after Combined Internal Carotid Artery Occlusion and Extracranial-Intracranial Anastomosis

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A patient with a giant aneurysm of the internal carotid artery (ICA) was treated by combined ICA occlusion in the neck and superficial temporal artery to middle cerebral artery anastomosis. Six months after complete closure of the ICA, the patient had episodes of nasal discharge mixed with dark red, old blood. Computed tomographic scan demonstrated that the lumen of the giant aneurysm was filled with a large amount of air, which communicated with the nasal cavity. Not finding a similar case in the literature, we named this entity "giant pneumoaneurysm." After direct operation, the giant pneumoaneurysm collapsed. This unusual complication may be due to a decrease of blood supply to the aneurysmal wall through the vasa vasorum. (*Neurosurgery* 21:394-398, 1987)

Key words: Carotid occlusion, Computed tomography, Extracranial to intracranial bypass, Giant aneurysm, Internal carotid artery, Vasa vasorum

In recent years, ligation of the internal carotid artery (ICA) with extracranial-intracranial (EC-IC) bypass has been used for the treatment of surgically inaccessible or technically difficult intracranial aneurysms (7, 10, 11, 13, 21). Ischemic complications (4, 8, 9) and growth or rupture (5, 10) of the aneurysm after carotid occlusion have sometimes been reported. We treated a patient with a huge ICA aneurysm by combined ICA occlusion and superficial temporal artery to middle cerebral artery (STA-MCA) anastomosis. Computed tomographic (CT) scan obtained 6 months after occlusion of the ICA in the neck, however, demonstrated that the aneurysmal lumen was filled with a large amount of air; there had been no episode suggestive of rupture. Not finding any similar case reported in the literature, we have named this entity "giant pneumoaneurysm." A report of this case is presented with a discussion of the mechanism involved in such an unusual complication.

## CASE REPORT

A 43-year-old man underwent left carotid angiography in 1970 because of visual disturbance of the left eye. The angiogram disclosed a giant aneurysm of the intracavernous portion of the ICA. No treatment was provided for the giant aneurysm at that time, and about 1 year later the patient became blind in the left eye. The course thereafter was uneventful except for slight mental disturbance with no clinical evidence of subarachnoid hemorrhage.

### First admission

For the purpose of reexamining the giant aneurysm, the patient was admitted on May 10, 1982. Neurological examination revealed the patient to be alert but slightly disoriented, with complete ophthalmoplegia on the left. He complained of impaired light touch and pain sensation on the left face. CT scans showed that there was a huge, partially calcified, globoid lesion of high density in the left middle cranial fossa, which eroded the basal bone structures including the sphenoidal ridge and the petrous pyramid (Fig. 1). The lesion measured 6 × 6 × 7 cm. After the administration of contrast medium, the eccentric and peripheral zones were intensely enhanced (Fig. 1, B and C). Left carotid angiography disclosed

a partially thrombosed giant aneurysm from the left ICA with elevation of the M<sub>1</sub> portion of the middle cerebral artery (MCA) by a large avascular mass (Fig. 2). Right carotid angiography demonstrated collateral flow to the left MCA from the right carotid artery. Otorhinolaryngological examination revealed bulging at the left upper corner of the pharynx due to the giant aneurysm. Endocrinological studies were essentially normal.

On August 24, 1982, the ICA was surgically exposed in the neck, and electroencephalographic (EEG) monitoring and regional cerebral blood flow (rCBF) measurements by the <sup>133</sup>Xe inhalation method during temporary ICA clamping were made. The patient's clinical condition and EEG pattern did not change during ICA occlusion for 30 minutes. Left rCBF was within the normal range (48 ml/100 g/minute) during ICA occlusion. The stump pressure with the ICA opened was 155 mm Hg and, with the ICA clamped, the pressure fell to 90 mm Hg. We partially ligated the ICA in the neck to allow the mean pressure to be 131 mm Hg, a midvalue between the opened and closed values, so that the transcranial pressure gradient could be enlarged and the flow through the graft could be enhanced (6).

On September 1, an STA-MCA anastomosis was performed, but a contusion was observed in the cortex of the temporal lobe. This was perhaps due to the use of an unsatisfactory skull perforator. We therefore hesitated to proceed with complete closure of the ICA in the neck. Left carotid angiography performed on October 29 and December 6 showed the aneurysmal lumen to be decreased. On December 9, the patient was discharged.

### Second admission

A follow-up carotid angiogram on September 19, 1983, demonstrated that the lumen had enlarged compared to that observed in the previous angiogram. On October 12, the ICA in the neck was exposed and a Selverstone clamp was applied, which was gradually closed over 7 days. Cerebral angiography conducted on November 25, however, showed the occlusion of the ICA to be incomplete. The ICA was still patent, but aneurysmal filling was absent except for a small portion of the orifice (Fig. 3A). The patient was discharged on November 30.

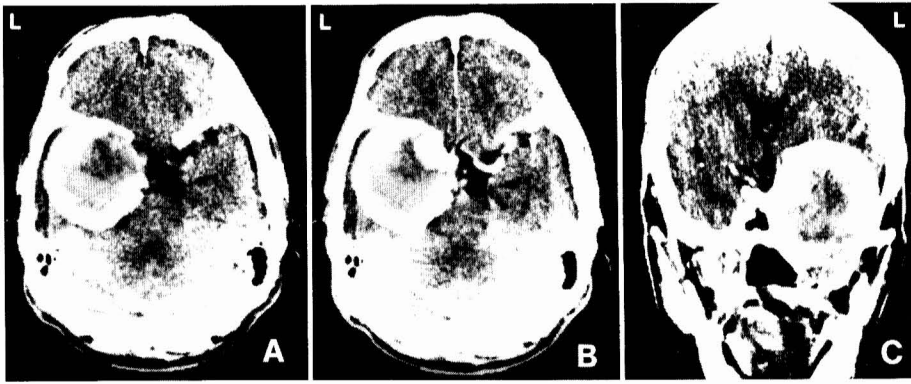


FIG. 1. CT scans obtained in 1982 at the first admission demonstrate a giant aneurysm measuring  $6 \times 6 \times 7$  cm. A plain scan (A) shows a spherical lesion with a partially calcified capsule occupying a substantial part of the left middle cranial fossa. Postcontrast scans (B and C) show enhancement of an eccentric area and the rim of the peripheral region. A coronal scan (C) shows that the basal bone structures are eroded.

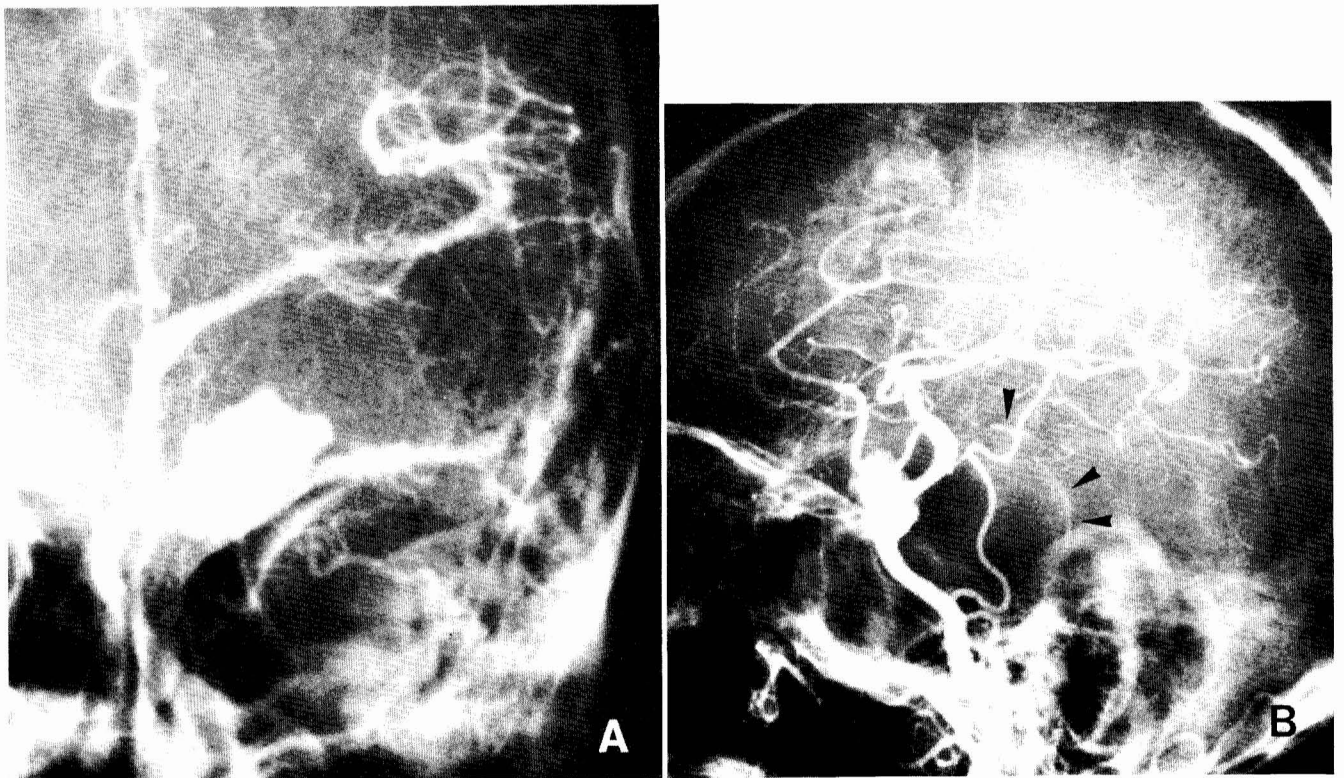


FIG. 2. Left carotid angiograms at first admission. A large, partially thrombosed aneurysm of the ICA is associated with upward displacement of the M<sub>1</sub> portion of the MCA (A). A shell-like calcification (arrowheads) is seen in the temporal region (B).

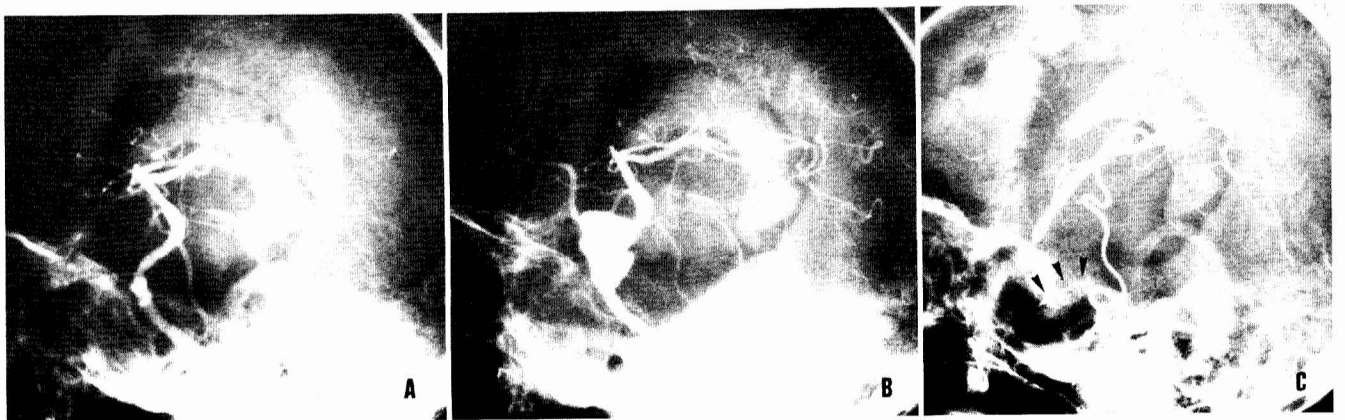


FIG. 3. Follow-up left carotid angiograms, lateral view. A, on November 25, 1983, the carotid system is still opacified because of incomplete occlusion of the ICA, but there is no aneurysmal filling except of a small portion of the orifice. B, on July 20, 1984, the aneurysmal lumen is larger than at the first admission 6 days before complete occlusion of the ICA. C, on March 15, 1985, MCA filling by the STA-MCA anastomosis and the linear calcification (arrowheads) has dropped downward 9 days after direct operation.

### Third admission

The patient's clinical course was uneventful. A postcontrast CT scan on July 20, 1984, showed that the eccentrically enhanced area had become obviously larger (Fig. 4A). Left carotid angiography performed on the same day revealed that the aneurysmal lumen had become larger than at the first admission (Fig. 3B). We therefore exposed the Selverstone clamp in the neck and completely closed it. Repeat CT scans with contrast enhancement showed that the attenuation values of the eccentric round part and the peripheral zone had decreased (Fig. 4B).

### Fourth admission

In January 1985, the patient began to complain of headache and of episodes of nasal discharge mixed with dark red, old blood. He was hospitalized on February 4, and CT scans at admission demonstrated that the giant aneurysmal cavity was filled with a large amount of air, which communicated with the nasal cavity (Figs. 4C and 5A). Examination of the region from the pharynx to the nasal cavity by an otolaryngologist failed to identify an opening to the giant aneurysm.

On March 6, 1985, with the intention of packing muscle pieces into the aneurysmal cavity, exploration of the mass was undertaken by left temporal craniotomy. When the dura mater was opened, the gyri of the anterior temporal lobe were necrotic and the sylvian fissure was elevated. A large, non-pulsatile mass covered by the dura mater of the middle cranial fossa was readily exposed. A 2-cm aneurysmotomy was performed. The aneurysm had been hollow, with the wall being 4 to 5 mm thick. There were old thrombus and pus within

the aneurysmal cavity, which we removed by suction and irrigation with amikacin solution. The internal side of the aneurysmal wall was yellowish white and smooth, and the orifice of the ICA could no longer be identified. There was a hole 8 mm in diameter leading to the upper pharynx. The aneurysmal wall was partially resected and then closed directly without packing.

Immediately after the operation, the patient's consciousness was clear, with no remarkable change except fever. On the 5th postoperative day, however, right hemiparesis developed and continued for about 1 month. Postoperative CT scans showed gradual dropping of the upper wall of the aneurysm (Fig. 5, B to D). Left carotid angiography also demonstrated downward deviation of the MCA branches, which filled via the STA-MCA anastomosis (Fig. 3C). A coronal CT scan performed on December 18, 1985, showed sinking of the upper wall of the aneurysm toward the middle cranial fossa (Fig. 5D). Histopathological examination of the aneurysmal wall obtained at operation revealed the presence of many blood vessels in the boundary zone where the dura mater is attached to the aneurysmal wall and in the inner and outer zones of the aneurysmal wall (Fig. 6). The patient was discharged on May 9, 1985, and 2 years after discharge was doing well.

## DISCUSSION

To our knowledge, there has been no report in the literature of retention of air within a giant aneurysm. We have named this entity "giant pneumoaneurysm." After aneurysmotomy

FIG. 4. Axial postcontrast CT scans before and after complete closure of the cervical ICA. A, CT scan 6 days before complete occlusion of the ICA shows that the eccentric enhanced area has become larger than at the first admission. B, CT scan 4 months after complete closure of the Selverstone clamp demonstrates that the attenuation values of the eccentric round part and the peripheral zone decreased with time. C, the aneurysmal cavity is filled with a large amount of air on February 4, 1985, 1 month before direct operation.

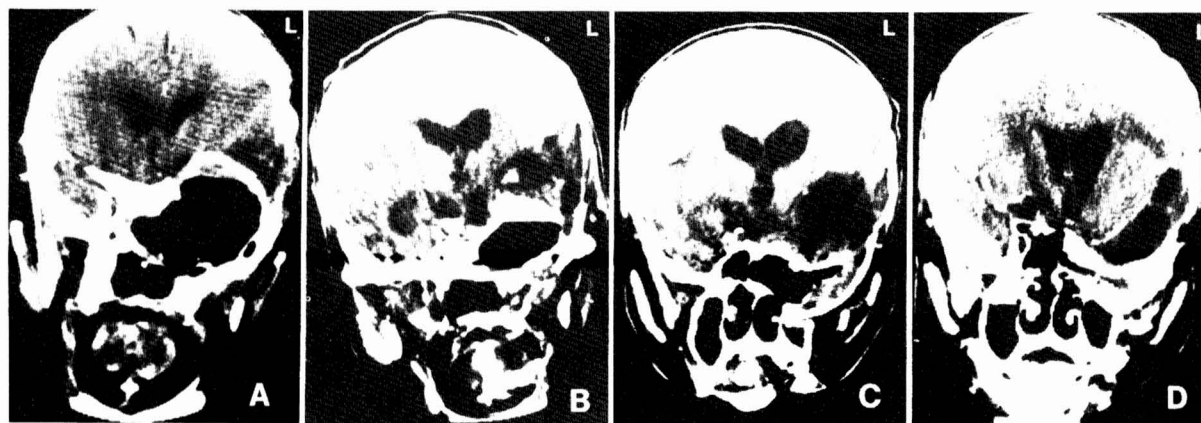
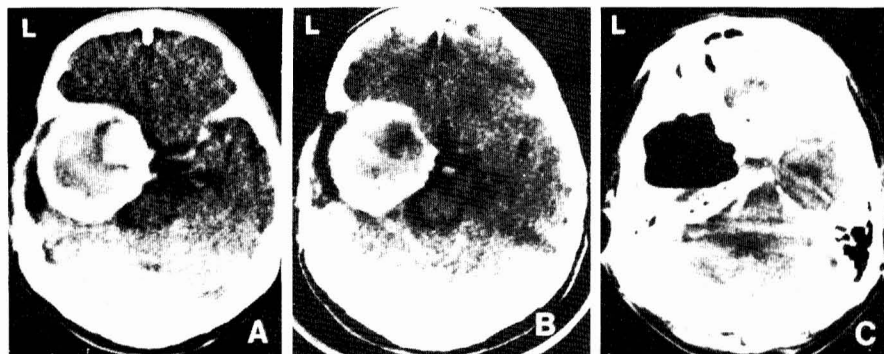


FIG. 5. Repeat coronal CT scans of the giant pneumoaneurysm. A and D are postcontrast scans, and B and C are plain scans. A CT scan on February 4, 1985, 1 month before direct operation, shows that the giant aneurysmal cavity filled with air is communicating with the nasal cavity (A). After direct operation, the upper wall of the aneurysm sank with lapse of time, leaving behind a hypodense area: B, the following day; C, 23 days later; D, 9 months later.

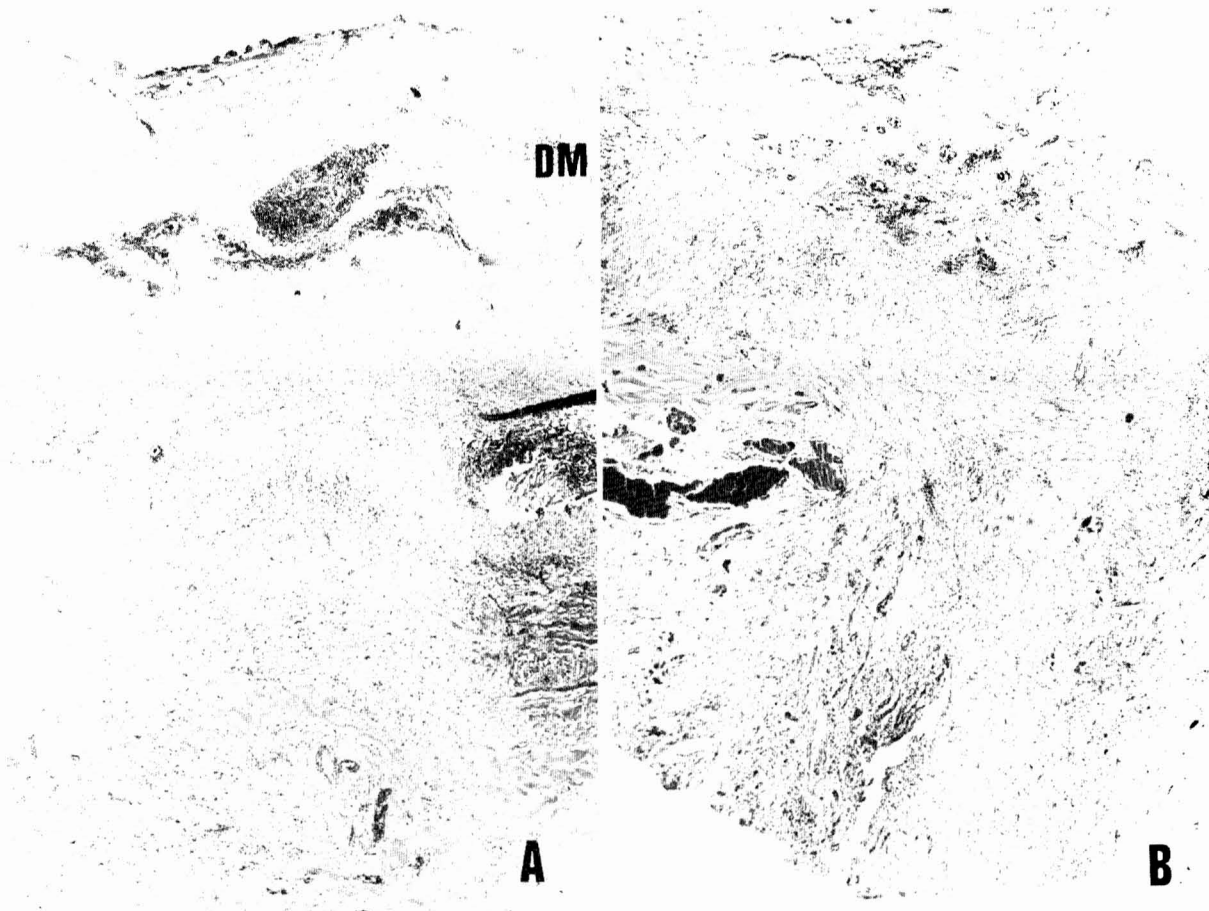


FIG. 6. Photomicrographs of a surgical specimen of the wall of the giant pneumoaneurysm, which can be roughly divided into three zones: vessel-rich inner and outer thirds and vessel-poor middle third. (Hematoxylin and eosin stain,  $\times 30$ .) *A*: Photomicrograph of the outer half of the aneurysmal wall covered with dura mater (*DM*). The outermost layer consists of dense fibrocollagenous tissue of dura mater firmly attached to the adventitial surface of the sac, and blood vessels are observed in this boundary zone. Many capillary spaces with rather prominent lymphocytic infiltrates can be seen in the outer third of the wall. *B*: Photomicrograph of the inner half of the wall shows granulation tissue with abundant capillaries and perivascular lymphocytic infiltrates. In the *right lower corner* can be seen many tiny clefts of cholesterol crystals.

for giant pneumoaneurysm, the patient became hemiparetic. In view of the findings of angiography and CT scanning, it is presumed that the upper wall of the hollowed aneurysm sank into the middle cranial fossa, followed secondarily by traction downward of the lenticulostriate arteries, thus leading to hemiparesis. After incomplete occlusion of the ICA, aneurysmal rupture did not develop even though the aneurysmal lumen was enlarged compared to the size observed on admission. After complete occlusion of the ICA, an opening formed in the aneurysmal wall that would have produced a massive hemorrhage had there been blood flow. We think that the giant aneurysm thrombosed after complete occlusion of the ICA, the propagation of thrombosis occluded the aneurysm's parent artery, the peripheral part of the aneurysm including the neck became organized, the wall became ischemic, necrosis developed in the weakest part of the ischemic wall to form an opening, the unorganized thrombus flowed into the nasopharynx, and then air filled the hollowed aneurysmal cavity.

Swearingen and Heros described a patient who died from rupture of a giant basilar artery aneurysm that had previously been demonstrated to have thrombosed spontaneously and completely (24). As the possible cause of rupture of a completely thrombosed aneurysm, they proposed rupture through the tiny portion of the neck that remained unthrombosed or rupture after recanalization of the aneurysm. Matsuda et al.

presented a case of fatal hemorrhage that occurred from a previously unruptured giant ICA aneurysm after thrombus formation within the aneurysmal sac, which had been confirmed by angiography and CT scanning after ICA occlusion with EC-IC bypass (13). In their case, as the small residual lumen was opacified near the aneurysmal neck, they attributed the rupture of the aneurysm to increase in retrograde flow through the bypass, resulting in reexpansion of the aneurysm. Various reports in the literature, however, have shown that carotid ligation not only lowers the incidence of aneurysmal rupture, but also reduces the pressure in the carotid artery, with a resultant decrease in the size of ICA aneurysms (or thrombosis) in a majority of cases (5, 7, 8, 10-12, 14, 19, 21). Most STA-MCA anastomoses with a flow of only 20 to 60 ml/minute at the time of operation cannot replace all of the blood flow of the parent artery (16, 21). There is thus an obvious decrease in blood flow after ICA occlusion with STA-MCA bypass. It is, therefore, difficult to conceive that a completely thrombosed giant aneurysm after operation would rupture only from a change of blood flow. In the case of Matsuda et al., as in our case, CT scans after carotid occlusion showed an almost typical appearance (13). In fact, autopsy of their patient showed the aneurysm to be filled with thrombus, leaving a narrow residual lumen.

In partially or completely thrombosed giant aneurysms,

ring-shaped contrast enhancement of the aneurysmal wall or shell-like calcification may be observed on CT scans. The former characteristic is deemed to have resulted from enhancement of the vascularized wall (1, 12, 17-20, 22, 25). Usually, after carotid occlusion, plain CT scans show an increase in attenuation values of the aneurysmal lumen, reflecting thrombosis immediately after operation. Thereafter, the aneurysm gradually loses its well-defined margin and the density continues to decrease from the periphery to the center of the aneurysm, but no enhancement is noted after contrast infusion during this course (12, 13, 19, 20).

During histological examination of the giant aneurysmal specimens obtained at operation or necropsy, many small vessels resembling vasa vasorum are observed in the aneurysmal wall (3, 13, 18, 20, 22, 23, 26, 27). It is reasonable to assume that the thickened wall of giant aneurysms requires vasa vasorum to provide nourishment. Pinto et al. postulated that the thickened fibrous wall surrounding the aneurysmal lumen containing many small vessels is formed from multipotential cells of the adjacent meninges in response to the subarachnoid mass (18). Mitsugi et al. demonstrated that, even after neck clipping without aneurysmectomy, the wall of a giant aneurysm was enhanced on a postcontrast scan, presumably due to a meningeal reaction (15).

We think that, after decrease or disappearance of blood flow in the aneurysmal lumen, the blood supply to the vasa vasorum of the wall, in particular the blood supply to the internal side of the wall, decreases, leading to ischemia of the wall (depletion of ring-shaped enhancement was observed on CT scans). Hemodynamic injury through the recanalized channel or the residual lumen aggravates a weak point in the wall and, if damage to the wall is significant, the aneurysm ruptures. If ischemic change in the aneurysmal wall is severe, a hole may develop in the wall even though a hemodynamic injury does not occur. Whenever a residual lumen is observed in the postoperative angiogram after the proximal occlusion of giant aneurysms, combined intracranial trapping of the aneurysm should be considered (2, 21).

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