Giant serpentine intracranial aneurysm after carotid ligation

Case report

HARALD FODSTAD, M.D., BENGT LILIEQUIST, M.D., STAFFAN WIRELL, M.D., PER-ERIK NILSSON, M.D., LENNART BOQUIST, M.D., AND ALI ABDUL-RAHMAN, M.D.
Departments of Neurosurgery, Neuroradiology, and Pathology, University of Umeå, Umeå, Sweden

A case is reported of a giant aneurysm of the intracavernous portion of the left internal carotid artery that was treated initially with a left common carotid artery ligation. Six months later the aneurysm was partially removed. During this time the development and evolution of thrombus formation, a serpentine channel, and a hyper-vascular capsule was easily followed with repeated computerized tomography and angiography of the aneurysm.

KEY WORDS: giant aneurysm, serpentine channel, angiography, computerized tomography, Coanda effect, blood flow

INTRACRANIAL aneurysms over 25 mm in diameter are rare. Although these giant aneurysms commonly originate from the subclinoid portion of the internal carotid artery (ICA), they are also found on the supraclinoid ICA, middle cerebral artery, basilar artery, and peripheral arterial branches in the posterior cranial fossa. The appearance of a spontaneous partial thrombosis with a serpentine channel through the aneurysm is unusual. The following is a report of such a case.

Case Report

Since early childhood, this 27-year-old woman had had a ptosis of the left eye. During pregnancy in 1969 she complained of an intense headache; examination revealed a partial third nerve palsy on the left side. X-ray examination of the skull after head trauma in 1973 showed destruction of the floor of the left middle cranial fossa and the left side of the clivus. Repeat clinical examination revealed partial paresis of the left third and sixth cranial nerves. In December, 1976, the patient developed increasing pain behind the left eye, left hemifacial paresthesia, and diplopia.

EXAMINATION. On admission the blood pressure was normal. Hyposmia, exophthalmos with normal fundus, and a central scotoma were present on the left side. Vision was blurred in the left eye, and there was a partial left oculomotor and abducent nerve paresis. The corneal reflex was weaker on the left side. Left hemifacial hypesthesia and hypalgesia were found.

An electroencephalogram (EEG) revealed a left frontotemporal abnormality. X-ray
FIG. 1. Plain x-ray film (upper) and computerized tomography scan (lower) performed on admission, showing destruction of the left middle cranial fossa.

Fig. 2. Admission computerized tomography scans before (left) and after (right) contrast enhancement.

study of the skull and orbits, and computerized tomography (CT) scan showed a partial destruction of the base of the middle cranial fossa, the posterior clinoid process, and adjacent part of the clivus and left sphenoid wing (Fig. 1). A circumscribed expanding mass in the left middle fossa enhancing with intravenous contrast injection was seen on CT scanning (Fig. 2). Selective angiography of both ICA's and the left vertebral artery showed a giant aneurysm originating from the extradural, intracavernous portion of the left ICA (Fig. 3). The aneurysm was filled by a jet of contrast material from behind and medially. The left hemisphere was supplied with blood from the basilar artery via the left posterior communicating artery as well as from the giant aneurysm. The largest diameter of the aneurysm was 7.5 cm, and the volume was calculated to be 100 cu cm.7

Course. On January 12, 1977, a Seldinger clamp was applied to the left common carotid artery. Two days after complete vessel occlusion, the patient complained of progressive diplopia and developed a total left ophthalmoplegia. Ophthalmodynamometry showed a significant lowering of the systolic pressure in the left eye compared to the preoperative study, and an EEG demonstrated that the left-sided abnormality had increased. A CT scan 3 days after total occlusion showed that the aneurysm had probably diminished slightly in size (Fig. 4). The attenuation values had increased markedly in the central part of the aneurysm with only minimal increase after enhancement with contrast material.
Giant serpentine aneurysm

Two months later the patient complained of increasing left-sided headache. Her neurological status was unchanged except for left optic atrophy. During aortocervical angiography the aneurysm appeared only as a serpentine channel. Enlarged extracranial anastomosis between the left vertebral artery and the left occipital artery filled in a retrograde direction the left ICA and the aneurysm (Fig. 5). The CT scan showed that the aneurysm remained the same size; however, there was a marked enhancement of a thin capsule surrounding an area of low attenuation values, probably representing an organized thrombus. In the medial part of this was a region with increased attenuation values (increased density) corresponding to the serpentine passage of blood through the aneurysm (Fig. 6). An increasing thickness of the capsule but no increase in size of the mass was demonstrated by CT scan 1 month later.

At the end of June, 1977, the patient suffered progressive headache with nausea and vomiting. A repeat CT scan on July 11 showed the appearance and size of aneurysm unchanged. Right carotid, left vertebral, and selective left carotid angiography with percutaneous puncture of the ICA distal to the Selverstone clamp demonstrated a tortuous serpentine vascular channel through the aneurysm with passage of blood to the left carotid siphon, left ophthalmic artery, anterior cerebral artery, and middle cerebral artery (Fig. 7). There were two openings into the aneurysm: one posteromedially from the ICA and one anteriorly into the carotid siphon.

Operation. On July 27, 1977, the left ICA was ligated in the neck and a left frontotemporal flap was turned. A temporal cricicotomy was performed and the aneurysm wall was found at a depth of 5 mm. The
A thickened wall was opened and large masses of organized hematoma were evacuated. Rather heavy bleeding from the opening deep inside the aneurysm was controlled with Heifetz clips permanently applied to the left ophthalmic artery and to the ICA proximal to the origin of the posterior communicating artery. The lateral part of the aneurysm wall was removed and the rest was sutured to the dural sheath.

**Pathological Examination.** Light microscopic study of the specimen showed partial thrombosis of the aneurysm with organization in the peripheral portions of the thrombus. Small, newly formed blood vessels lined by endothelial cells were seen in the thrombus (Fig. 8 upper). The thrombus also contained a large blood channel lined partly by endothelial cells and partly by granulation tissue (Fig. 8 lower). The wall of the aneurysm con-
Giant serpentine aneurysm


**FIG. 9.** Photomicrograph of a portion of the aneurysmal wall demonstrating hemorrhages and small blood vessels situated both in superficial and deep portions of the wall. H & E, × 80.
sisted of thick fibrous tissue with occasional fragments of elastic fibers, edema, and areas of hemorrhage. Lymphocytes, plasma cells, and a few granulocytes were found, usually close to the hemorrhagic sites. Small vessels with endothelial lining were found in the aneurysmal wall (Fig. 9). Calcification was not observed. The aneurysm was adherent to the dura, which showed fibrous thickening and areas of bleeding and granulation tissue.

Postoperative Course. The postoperative course was uneventful. A CT scan 5 days after operation showed increased attenuation values corresponding to that part of the aneurysm capsule that was left behind. A final CT scan without contrast enhancement 2½ months later showed no signs of the aneurysm. Six months postoperatively the patient's headache had disappeared. She showed only a partial left ophthalmoplegia and left fifth nerve paresis.

Discussion

The sequence of unusual changes in this type of giant aneurysm has not previously been studied by repeated CT scan and simultaneously repeated cerebral angiography. Giant aneurysms appear on CT scan as a mass lesion with attenuation values slightly above the surrounding soft tissues if it contains circulating blood, or with markedly increased values when it is partially or totally thrombosed. The increase in attenuation after contrast enhancement, and the shape and site of the aneurysm could occasionally cause difficulties in differentiating the mass from a meningioma or a neurinoma.

The finding of a thick capsule with vessels has been reported in a surgically removed giant aneurysm and the ring shadow appearing after contrast enhancement apparently corresponds with such a thickened vascular wall. At surgery, the capsule was found to be rich in small vessels from which profuse bleeding issued. Microscopic examination confirmed that the capsule consisted of a thickened and vascularized wall close to the dural sheet (Fig. 9). In our case there was originally no evidence of a capsule; the capsule was formed within 1 month of the formation of the thrombus. The cause for the formation of the capsule may be the thrombus itself, which stimulates the original thin wall to take part in the organization of the thrombus.

A ring shadow appearing after contrast enhancement during CT scanning has been reported in two patients with a giant aneurysm. These cases had no serpentine channel, and the aneurysms presented at angiography with a cavity diameter of only 5 mm and 7 mm, respectively. These authors theorize that the ring appearance on the CT scan is due to a presumed central clot surrounded by blood flow peripherally.

The following explanation for the appearance of a serpentine channel seen at angiography has been put forward by other investigators. The reduced velocity of the blood stream through the aneurysm caused by the common carotid artery occlusion in our case favors the formation of a thrombus. Spontaneous thrombosis with serpentine channel formation more often forms in giant aneurysms originating from the middle cerebral artery, although at least two such aneurysms arising from the carotid siphon are on record. This rare occurrence in giant ICA aneurysms may be due to two possibilities. First, there is a greater jet force of the blood stream entering the aneurysm directly from the carotid artery, thus preventing the blood from stagnating within the aneurysm. Second, subclinoid aneurysms are invested by a dural sheet and grow close to bony structures. Giant aneurysms originating from the middle cerebral artery, on the other hand, are not similarly buttressed and hence expand more easily. A partial thrombosis then occurs when the aneurysm has enlarged too much in relation to its orifice.

An explanation for the formation of a serpentine passage through the originally globular aneurysm may be the Coanda effect, in which the direction of a jet stream determines the direction of the blood flow (in this case along the wall of a large saccular aneurysm). Robinson and Roberts describe how fluid-pressure changes along the jet stream reinforce its pathway close to one wall of the disturbed artery. This theory tallies with the observation that hitherto published cases show similar angiographic pictures with the serpentine channel situated peripherally in the aneurysm.

We have located 12 cases in the literature in which angiography revealed serpentine channels inside giant aneurysms. Their bizarre angiographic appearance and unusual clinical behavior set them aside as an
Giant serpentine aneurysm

is an interesting subgroup of aneurysms. They seldom give rise to subarachnoid bleeding and usually appear as a mass lesion or cause signs of ischemia. As in our patient, all reported cases with a giant serpentine aneurysm had a history of 2 or 3 months of increasing headache during the process of apparent thrombus formation. The possibility also exists that a richly vascularized capsule could bleed repeatedly, causing enlargement of the aneurysm with subsequent clinical symptoms.

References
6. Jones JN, Schwarz HJ: Two cases of giant intracerebral aneurysm simulating neoplasm on CT scan; one with coexistent chronic subdural hematoma J Neurol 215:49-57, 1977

Address reprint requests to: Harald Fodstad, M.D., Department of Neurosurgery, Umeå University Hospital, 5-901 85 Umeå, Sweden.