Spontaneous carotid-cavernous fistula produced by ruptured aneurysm of the meningo-hypophyseal branch of the internal carotid artery

Case report

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The authors report a case in which the cause of a "spontaneous" carotid-cavernous fistula could not be demonstrated by selective angiography of external and internal carotid arteries. However, postmortem study of the cavernous sinuses revealed an unsuspected ruptured aneurysm of the internal carotid artery at the origin of the meningo-hypophyseal branch.

KEY WORDS: carotid-cavernous fistula, intracavernous carotid aneurysm, meningo-hypophyseal artery

In a review of internal carotid-cavernous fistulas (CCF), 10% to 25% have been categorized as "spontaneous." According to Newton and Hoyt, most of these fistulas are produced by shunts between meningeal branches of the internal and external carotid arteries and dural veins in the vicinity of the cavernous sinus. Aneurysms in the cavernous portion of the internal carotid artery represent from 3% to 11% of the total number of intracranial arterial aneurysms. However, the majority of carotid aneurysms do not produce a CCF and only a few related observations have been made.

The ruptured aneurysms inside the cavernous sinus are usually small since the larger ones obliterate the cavernous region and act as an expansive extradural or subclinoidal mass. Their origin may be congenital or more often arteriosclerotic. The development of a significant CCF may prevent the recognition of a small aneurysm in routine angiographic studies. Dandy and Hamby have deplored the lack of well-documented reports of postmortem examinations in cases of CCF; in fact, Hamby remarked that the best illustrations of these lesions are still those published by Delens in
his thesis of 1870. We are reporting the postmortem study of such a case.

**Case Report**

Five months before admission this 46-year-old woman noticed right frontal headaches of increasing intensity. Shortly afterward, she became aware of a noise in the right side of her head, and 2 months later the onset of progressive right exophthalmos, diplopia, and loss of vision on the right side. She had no previous history of trauma or related disease.

**Examination.** There was a non-pulsating right exophthalmos with 23 mm protrusion as compared to 16 mm in the left eye. Right visual acuity was 0.5. The right eyelid was edematous. The retinal veins were enlarged and there was some blurring of the optic disc in the right fundus. Diplopia was experienced when the patient looked to the right and upward. A continuous bruit synchronous with the pulse was heard over the right fronto-orbital region; the phonocardiogram showed the bruit during systole and with a higher amplitude over the right orbital and frontotemporal region. When the right carotid was compressed in the neck, the bruit was neither heard nor recorded.

The rest of the neurological and cardiovascular examination was negative. Compression of the left carotid artery seemed to induce slower electroencephalographic activity.

Angiographic injection of the right internal carotid artery demonstrated early filling of some small veins in the cavernous region that seemed to correspond to superior and inferior components of the cavernous venous plexus (Fig. 1 left). Later, there was massive filling of the cavernous sinus with drainage toward the right superior and inferior ophthalmic veins and the inferior petrosal sinus (Fig. 1 right). Injection of the right external carotid artery revealed several arterial branches of the internal maxillary, middle meningeal and ascending pharyngeal arteries that reached the cavernous sinus and afterward the superior ophthalmic vein. Angiography of the left internal carotid artery demonstrated intracavernous branches of the carotid, one of which could be identified as the meningohypophyseal artery (Fig. 2). Later the right superior ophthalmic vein was visualized. Injection of the left external carotid artery visualized a rich network of fine branches of the ascending pharyngeal, middle meningeal,

**Fig. 1.** Left: Right internal carotid angiogram, early arterial phase. Several small vessels (arrows) can be seen arising from the intracavernous portion of the internal carotid artery. Right: In a later phase there is filling of the cavernous sinus and some of its veins. The right superior ophthalmic vein (vertical arrow) and the right inferior petrosal sinus (horizontal arrow) are shown.
Aneurysm of meningohipophyseal artery

Fig. 2. Left internal carotid angiogram, demonstrating filling of small meningeal arteries arising from the cavernous portion of the internal carotid artery. Meningohypophyseal artery (upper arrow) and inferior cavernous sinus artery (lower arrow) are seen.

and internal maxillary arteries flowing toward the cavernous sinus; later the cavernous sinus, right superior ophthalmic vein, and inferior petrosal sinus filled.

Operations. In the first operation, both external carotid arteries were ligated. There was some reduction of the right exophthalmos and bruit, but this only lasted a few days.

One month later intracranial clipping of the right internal carotid artery was followed in the same operation by ligature and muscle embolization through the internal carotid artery in the neck. Unfortunately, the intracranial silver clip gave way, allowing one of the pieces of muscle to ascend and induce thrombosis of the internal carotid and middle cerebral arteries. The patient died 2 days later.

Postmortem Examination. At autopsy a large block was removed containing the body of the sphenoid bone, sella turcica, anterior part of the circle of Willis, the optic nerves and chiasm. A careful anatomical study was then carried out with the help of the surgical microscope. The right carotid and middle cerebral arteries were completely thrombosed. There was an anomaly of the anterior part of the circle of Willis; both anterior cerebral arteries originated from the left carotid, and the anterior communicating artery was hypoplastic. The lateral wall of the right cavernous sinus was resected. The cavity of the sinus was filled with coagulated blood and a hard thrombus since the specimen had been fixed in formalin; for these reasons the dissection was difficult and some small intracavernous branches were damaged. In the posteroinferior portion of the intracavernous artery a small, round, ruptured aneurysm, 3 to 4 mm in diameter, was found at the point of origin of the meningohipophyseal branch. The aneurysm was surrounded by many widely dilated venous channels inside the cavernous cavity (Fig. 3).

Discussion

This postmortem observation of an unsuspected and ruptured aneurysm at the origin of the small meningohipophyseal branch of the intracavernous portion of the carotid artery may have relevance to some of the so-called "spontaneous" carotid cavernous fistulas (CCF). Moreover, the available anatomical knowledge concerning small arterial branches at the base of the skull within the cavernous sinus, has been expanded by several recent studies using refined radio-anatomical methods and demonstrating these arteries in normal angiograms. The exact point of origin of the CCF could be established radiologically in 24% of 50 cases studied by Djindjian, et al.

It seems also that the meningohipophyseal branch of the intracavernous carotid may be recognized in all normal angiograms using the refinements of present neuroradiological techniques, because this artery is a very constant branch as shown in the anatomical dissections. The rupture of this artery, which arises at the posterior part of the first curve of the internal carotid in its intracavernous course, is responsible for the development of posttraumatic CCF in more than half of the cases in which the origin of the fistula could be determined.

Cases like the one we have reported are particularly important because of the availability of Parkinson's approach for repair of the fistula while preserving the carotid
circulation,\textsuperscript{4,6} as well as the technique of applying local catheters to occlude the site of the fistula.\textsuperscript{2,15,19} There seems to be no doubt that methods such as ultrarapid serial angiography, subtraction, and magnification of the radiological images will soon determine the cause and location of many of these "spontaneous" fistulas and provide the background for their more rational and direct treatment.

**References**

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