Ruptured berry aneurysm of the anterior ethmoidal artery associated with bilateral spontaneous internal carotid artery occlusion in the neck

Case report

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A unique case is described of subarachnoid hemorrhage from a ruptured berry aneurysm of the right anterior ethmoidal artery, and its pathogenesis is discussed. The literature suggests an increased incidence of posterior circulation aneurysms in moyamoya disease, of contralateral anterior circulation aneurysms in agenesis of the carotid artery, and of aneurysms at various sites in spontaneous and iatrogenically acquired obstructions of the internal carotid artery. In addition to congenital anomalies of the vessel wall and systemic hypertension, increased regional blood flow should be regarded as an important factor in the generation of berry aneurysms.

KEY WORDS • cerebral aneurysm • internal carotid artery • anterior ethmoidal artery • carotid artery occlusion • subarachnoid hemorrhage • anastomotic circulation

The occurrence of berry aneurysms has been linked to congenital defects in the vessel wall, particularly affecting certain bifurcation sites within the intracranial arterial vasculature. Arterial hypertension is considered to be a major factor in the pathogenesis of berry aneurysms, and these lesions are unusually prevalent in patients with coarctation of the aorta, polycystic kidneys, and certain vasculopathies.

A case history is presented of subarachnoid hemorrhage (SAH) from rupture of a berry aneurysm at an apparently unique site, the right anterior ethmoidal artery. This case suggests an additional etiological factor, namely, increased regional blood flow.

Case Report

This 65-year-old man was admitted to the neurosurgical unit of the Toronto General Hospital on August 18, 1977, having suffered the sudden onset of severe headache 3 weeks before. He was said to have suffered from left hemiparesis 4 years before, which had resolved.

Examination. On admission, examination revealed a hypertensive confused man with neck stiffness and slight right hemiparesis. Four-vessel angiography showed a berry aneurysm of the right ethmoidal artery on the anterior fossa floor near the midline (Fig. 1), occlusion of both internal carotid arteries (ICA's) in the neck (Fig. 2), stenosis of the right vertebral artery at its origin, compensatory hypertrophy of the left vertebral artery, and a rich anastomotic flow through both external carotid and ophthalmic arteries (Fig. 1 right). With supportive treatment and bed rest, the patient's mental state returned to normal, but his right hemiparesis persisted.

Operation. The aneurysm was exposed through a right frontal craniotomy on August 29, 1977. It was embedded in a small clot which formed a cast of its fundus (Fig. 3). The aneurysm was obliterated by cautery since it was too adherent to the dura to be clipped. The patient recovered without incident, and was able to walk on his own despite his persisting right hemiparesis. He returned home on September 11, 1977. His neurological status remained unchanged at the time of his latest follow-up examination on November 8, 1977.
FIG. 1. Right common carotid angiograms. **Left:** Anteroposterior view showing filling restricted to the external circulation. A berry aneurysm (arrow) is situated to the right of midline on the anterior fossa floor. **Right:** Subtraction of the lateral view showing in addition a rich anastomotic flow from the external carotid circulation through the anterior ethmoidal branches into the ophthalmic artery. The aneurysm (arrow) is seen to arise in a branch of the right anterior ethmoidal artery.

Discussion

The only comparable case of which the author is aware is that reported by Mujica, et al., with SAH from one of two berry aneurysms at the point of dural penetration of an ascending pharyngeal artery. Although this artery is also known to be directly involved in extra-intracranial anastomoses, the patient apparently suffered from no other vascular abnormality.

The present patient had bilateral occlusion of the ICA's at their origins; one occlusion probably occurred at the time of his left-sided hemiparesis 4 years before. In compensation, the patient had developed bilateral extensive anastomotic connections between the external carotid circulation and the branches of the ophthalmic arteries, particularly the anterior ethmoidal artery. On the right side, a berry aneurysm had arisen in the anterior ethmoidal distribution, at the point of dural perforation, and was the cause of SAH. It is suggested that the increased blood flow through the anterior ethmoidal arteries precipitated berry aneurysm formation, exposing this artery, which is not normally subject to aneurysm formation, to the same forces that are at work at conventional aneurysm sites.

Such a concept is supported from four sets of observations. First, it has been observed that moyamoya disease is frequently accompanied by aneurysm formation in the distribution of the posterior choroidal and basilar arteries. Kodama, et al., studied 56 cases of moyamoya disease, and found three posterior cho-

roidal and two basilar aneurysms. They considered the former to be false aneurysms and the latter true berry aneurysms attributable to increased regional blood flow. Nagamine, et al., reviewing the literature, found 24 reported cases of moyamoya disease associated with true berry aneurysms, including their own. Thirteen (43.3%) of these aneurysms were in the posterior circulation, compared with an expected incidence of 5.3% to 9.6% in the general population. Aneurysm formation in moyamoya disease was attributed to increased blood flow in vessels still patent after carotid occlusion.

Second, there is a recognized association between anomalies of the cerebral vascular tree and aneurysm formation, usually considered to support the notion that the aneurysms, too, were the result of vessel wall anomalies. In some patients, however, the alteration of the hemodynamics as the result of the anomalies is an equally attractive explanation. Katz, et al., drew attention to the increased incidence of anatomical anomalies in the circle of Willis and/or of persistent carotid-basilar anastomoses in patients with anterior communicating artery berry aneurysms. They reported a unique case of an anterior communicating artery aneurysm associated with bilateral basilar-middle meningeal artery anastomoses. Kirgis, et al., also emphasized the association between aneurysms of the anterior communicating artery and gross anomalies of the circle of Willis. Stehbens and Cohen and Kristensen discussed the correlation of aneurysms with vascular anomalies of cerebral arteries. Spallone and Cantore³⁵
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found the incidence of kinks and coils in the extracranial ICA significantly higher in 76 cases of single supratentorial berry aneurysms than in 120 patients subjected to angiography who did not suffer from cerebrovascular occlusive disease or aneurysms. The incidence of stenosis of the artery was not significantly different in the two groups, however. The abnormalities in aneurysm patients were commoner on the side contralateral to the aneurysm. They believed that the arterial abnormalities increased "hemodynamic stress on the side opposite to the carotid lesion." Wissinger, et al., reported a case of basilar artery aneurysm associated with absence of a vertebral artery.

The most striking association of this kind is that of aneurysms with congenital absence of the ICA, a condition in which intracranial hemodynamics are clearly rearranged. Servo, reported a case of a right carotid syphon aneurysm with associated agenesis of the left ICA, and was able to find 35 reported cases of absence of the ICA, four of which were associated with contralateral ICA aneurysms, four with anterior communicating artery aneurysms, one with a middle cerebral artery aneurysm, and three with basilar artery aneurysms. Moyses described a case of basilar artery aneurysm in a patient with agenesis of the left ICA. He drew attention to the association of anterior communicating artery aneurysms with hypoplasia or absence of the proximal anterior cerebral artery, mentioning 12 instances out of 47 personal cases of aneurysm at that site. Burmester and Stender reported two cases of aneurysm ipsilateral to an aplastic ICA, and Lhermitte, et al., Lagarde, et al., Turnbull, and Turnbull described anterior communicating artery aneurysms associated with agenesis of the ICA. Rosen, et al., reported a basilar artery aneurysm and Tangchaw and Khaoborisut a contralateral middle cerebral artery aneurysm associated with the absence of a carotid artery. Such observations underline the importance of hemodynamic forces applied to the apex of an arterial bifurcation in aneurysm formation, as demonstrated by Ferguson.

Third, although more difficult to marshal (presumably because of the shorter interval during which abnormal hemodynamics are at work), evidence exists that acquired carotid artery occlusion also may favor aneurysmal formation, enlargement, or rupture. Portnoy and Avellanosa reported a right ICA aneurysm at the origin of the posterior communicating artery in a patient with left ICA stenosis in the neck, and culled nine other cases of carotid artery stenosis or occlusion associated with aneurysm from the English literature, stressing the "hydraulic" implications. Stern, et al., reported 15 cases of symptomatic carotid artery stenosis associated with asymptomatic berry aneurysms, all in the anterior circulation, and five cases of SAH associated with asymptomatic carotid stenosis. Of the former 15 cases, the aneurysms were ipsilateral to the stenosis in nine and contralateral in two, and stenosis was bilateral in four. Of the latter five cases, the symptomatic lesions were contralateral in two and ipsilateral in three; aneurysms were bilateral in two. Denton and Gutmann reported a case of asymptomatic middle cerebral artery aneurysm associated with ipsilateral carotid stenosis in the neck, and Fields and Weibel reported a case of anterior communicating artery aneurysm associated with ipsilateral carotid stenosis in the neck.
described two cases of intracranial aneurysm associated with ipsilateral carotid stenosis. Jaffe and McHenry\textsuperscript{14} reported a right ICA aneurysm associated with left ICA occlusion in the neck. Pool and Potts\textsuperscript{27} described a left ICA aneurysm associated with right ICA stenosis in the neck, Somach and Shenkin\textsuperscript{34} treated a right-sided intracerebral aneurysm associated with left ICA stenosis in the neck, and Shoumaker, et al.,\textsuperscript{33} documented a case of multiple intracranial aneurysms associated with carotid stenosis. These reports exemplify the well known association of aneurysm formation with acquired obstruction of the ICA in the neck, which presents a major problem in management.\textsuperscript{1} Although it can be argued that aneurysms and carotid stenosis, both being common diseases, will inevitably be associated in a certain number of patients by chance, nevertheless in the light of the other evidence, hemodynamic principles must still be considered.

Finally, association of aneurysms with iatrogenic carotid artery occlusion in the neck is of interest. Since carotid ligation is usually performed to treat known aneurysms, postoperative aneurysmal development is more difficult to evaluate.\textsuperscript{28} Hassler\textsuperscript{13} produced aneurysms in rabbits secondary to carotid occlusion. German and Black\textsuperscript{8} reported two patients, one dying of an apparently new aneurysm of the anterior communicating artery and the other of a contralateral ICA aneurysm, after carotid artery ligation in the neck for carotid-cavernous fistula and right cavernous carotid aneurysm, respectively. They also mentioned another patient with rupture of a posterior communicating artery aneurysm contralateral to a carotid artery ligated in the neck, stressing the hemodynamic implications. Miller, et al., (in a presentation at the Annual Meeting of the American Association of Neurological Surgeons, April, 1981) alluded to the more rapid development of aneurysms contralateral to a ligated carotid artery, and Hunt (personal communication, 1981) treated a patient whose aneurysm expanded and ruptured 5 to 6 years after carotid ligation, associated with strong contralateral flow from posterior and anterior communicating arteries. Salar and Mingrino\textsuperscript{11} have reported that, among 126 patients treated with cervical carotid ligation, two went on to develop new aneurysms not present at the time of initial angiography. One occurred 9 years after right ICA ligation for a right-sided aneurysm in the siphon of the left supraclinoid carotid, and the other 15 years after right common carotid artery ligation for a right-sided aneurysm at the origin of the left posterior communicating artery. These authors felt that aging, arteriosclerosis, and altered blood flow were responsible. Somach and Shenkin\textsuperscript{34} reported that two of six patients followed for 3 to 10 years after cervical carotid artery ligation out of a total of 20 patients developed apparently new aneurysms, one in the parasellar contralateral carotid artery and the other in the anterior communicating artery. They attributed the development of the aneurysms to hemodynamic disturbances. Winn, et al.,\textsuperscript{41} treated half of a group of 60 patients who had a single posterior communicating aneurysm with cervical common carotid artery ligation, and the other half conservatively. There were no instances of postoperative SAH in the conservatively treated group, but one of the surgical group developed a new posterior communicating artery aneurysm which bled on the contralateral side 3 years and 8 months later. Hardy, et al.,\textsuperscript{11} repeated angiograms after 2 months to 6 years in 10 of 54 patients undergoing cervical carotid ligation for intracranial aneurysm, and demonstrated two cases with previously unsuspected aneurysms. Vlahovitch, et al.,\textsuperscript{40} pointed out that although ligation of the ipsilateral carotid artery in the neck usually has a beneficial effect on certain intracranial aneurysms unsuited to direct attack, it may sometimes, as in one of their patients, be followed by enlargement of the aneurysm; or it may result in the development of a \textit{de novo} aneurysm, as in another of their patients 17 years after common carotid ligation that cured a giant posterior communicating artery aneurysm. Other authors have also expressed this concern.\textsuperscript{9,10,25,26,30} Thus, there is considerable evidence to suggest that longer term \textit{de novo} aneurysm generation may follow carotid ligation.

Although in most cases no controlled studies are possible, there appears to be an unusual association of basilar aneurysms with moyamoya disease, particularly of contralateral anterior circulation aneurysms with agenesis of the carotid artery, and of various intracranial aneurysms with both spontaneous and iatrogenically induced occlusion of the ICA. Although the presence of aneurysm in congenital absence of the carotid artery and the well recognized relationship with other vascular anomalies could be attributed to associated congenital anomalies of the vessel wall, the occurrence of these malformations in acquired carotid disease and in moyamoya disease suggests that abnormalities of hemodynamics, which the congenital lesions also produce, are at least an equally important factor as they were in the case reported here. It is suggested that increased regional blood flow is an important factor in the generation of cerebral aneurysms, along with congenital defects in the vessel wall, systemic hypertension, and certain other factors.\textsuperscript{5,12,13}

References


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