De novo formation and rupture of an azygos pericallosal artery aneurysm

Case report

WOLFGANG DIETRICH, M.D., ANDREA REINPRECHT, M.D., ANDREAS GRUBER, M.D., AND THOMAS ZECH, M.D.

Department of Neurosurgery, University of Vienna Medical School, Vienna, Austria

An azygos pericallosal artery (APCA) aneurysm is a rare anomaly that is closely associated with saccular aneurysms. This is the earliest report to document de novo formation and rupture of an aneurysm at the bifurcation of an unpaired pericallosal trunk. The authors report the case of a woman who presented at the age of 52 years with subarachnoid hemorrhage (SAH) from the rupture of a newly formed APCA bifurcation aneurysm, 7 years after she had undergone surgery to clip a ruptured anterior cerebral artery aneurysm. De novo formation of aneurysms after SAH rarely occurs and certain risk factors like multiple and familial aneurysms, arterial hypertension, or smoking have been postulated. Late follow-up examination with angiography to detect de novo aneurysms should be considered in patients with this vascular anomaly after SAH.

KEY WORDS • azygos pericallosal artery • de novo aneurysm • developing aneurysm

Case Report

History. This woman first presented to our clinic in 1985 at age 45 years after briefly losing consciousness with headache, neck stiffness, and drowsiness. The CT scan revealed an SAH with diffuse bleeding into the basal cisterns and the frontal interhemispheric fissure. Selective four-vessel cerebral angiography revealed an anomaly of the ACA complex, with an APCA and a right frontopolar artery branching off the A1 segment and an A1 aneurysm originating from this branching, with no evidence of additional aneurysms (Fig. 1). After the aneurysm had been successfully clipped, the patient recovered completely, but later suffered from arterial hypertension and was treated with antihypertension medication. Seven years later the patient suddenly collapsed without losing consciousness and was again admitted to our hospital.

Examination. At admission the patient presented with mild headache, neck stiffness, and mild cognitive impairment. A CT scan obtained at this time revealed blood in the frontal interhemispheric fissure and an intracerebral hematoma in the right frontal lobe. Selective four-vessel cerebral angiography results demonstrated an aneurysm with a diameter of approximately 1.3 cm at the bifurcation of the APCA (Fig. 2). There was no evidence of aneurysm recurrence at the site of previous aneurysm clipping. After the angiography study was completed, the patient’s head-
ache grew more intense and she became drowsy. A CT scan revealed an increase in the size of the intracerebral hematoma, with blood also located in the corpus callosum and the ventricles.

**Operation.** Immediately after the rebleeding episode, an interhemispheric approach was used to clip the aneurysm without requiring temporary clipping of the APCA.

**Postoperative Course.** The patient’s postoperative course was uncomplicated. Extubation could be performed on the 2nd postoperative day. No postoperative vasospasm occurred. Material placed for external ventricular drainage could be removed on the 5th postoperative day. The follow-up examination performed 2 months after surgery revealed chronic organic brain syndrome and chronic posthemorrhagic hydrocephalus. The patient was lost to further follow up.

**Discussion**

In this report we document the development of a saccular aneurysm at the bifurcation of an APCA trunk within 7 years after rupture and clipping of an A1 aneurysm. The newly formed aneurysm arose from an area that had appeared normal in angiography studies performed at the time of the first SAH and caused rebleeding by its rupture.

De novo formation of aneurysms has been reported to occur after CA ligation, and has been associated with the hemodynamic changes that result from this procedure.3,4 Because CA ligation has recently been abandoned as a routine therapeutic modality in aneurysm disease, however, the clinical significance of these reports is limited to special cases. De novo formation is rarely reported in cases in which this therapy has not been used.2,5,9,10,12 Patients with multiple aneurysms are reported to be at higher risk of rebleeding from de novo aneurysm formation.2,5,11 A genetic basis of the pathogenesis of arterial dysplasia in these patients is considered.

Miller and associates9 reported the formation of new aneurysms in sites previously observed to be normal in angiography studies in six patients after rupture and successful clipping of cerebral aneurysms. None of the patients had multiple aneurysms. The new aneurysms ruptured between 3 and 20 years after the original rupture. Similar time periods for the formation and rupture of a de novo aneurysm have been reported by other authors.7,10,12 Although the influence of arterial hypertension on the overall risk of SAH seems controversial, it appears to be an important risk factor for the development of new aneurysms in patients with SAH. Reviewing 49 cases of de novo aneurysm formation described in the literature,
Tonn and colleagues\textsuperscript{12} reported that arterial hypertension or smoking increases the overall risk of de novo aneurysm formation and that arterial hypertension also shortens the interval during which the aneurysm forms and ruptures. Arterial hypertension was also present in our case. In their report on de novo saccular aneurysms, Misra, et al.,\textsuperscript{10} discussed the contributory effect exerted by female sex hormones on aneurysm formation, inducing structural changes in the arterial wall. In their review, Tonn and colleagues\textsuperscript{12} could not document a female preponderance in de novo formation of aneurysms; nor has such a preponderance been documented in other series.\textsuperscript{5,9}

De novo formation of an aneurysm in an APCA has not been reported thus far. An APCA is a rare anomaly, but has clinical significance because of two reasons: it is associated with a high incidence of aneurysm formation, and, in the event of injury to the APCA trunk during an aneurysm clipping procedure, there will be deleterious results, including ischemia affecting both hemispheres. In 1965 Pool and Potts\textsuperscript{11} postulated a higher incidence of pericallosal artery aneurysms in patients with an APCA. Laitinen and Snellman,\textsuperscript{8} in a study of 14 patients with pericallosal artery aneurysms, found an APCA in three cases. Huber, et al.,\textsuperscript{6} in a study of the relationship between the frequency of unpaired pericallosal artery trunks and the incidence of saccular aneurysms of the pericallosal artery, found an APCA trunk bifurcation aneurysm in 41.1\% of cases. Of all pericallosal artery aneurysms in their angiographic material, 25\% were located at the bifurcation of an APCA trunk. So this bifurcation seems to have the highest incidence of aneurysms of any bifurcation of an intracranial artery.

Conclusions

This report adds new information in the quest to identify which patients who have experienced an SAH are at risk of developing new aneurysms. In their review Miller and associates\textsuperscript{9} calculated the risk of SAH in patients who had already undergone aneurysm surgery to be six times higher than those of the general population. Misra, et al.,\textsuperscript{10} in their report on de novo aneurysms in two women, suggested that follow-up angiography be performed, especially in patients with multiple aneurysms and in those who smoke, take oral contraceptives, or have arterial hypertension. Tonn and colleagues,\textsuperscript{12} in an investigation of risk factors for de novo formation of aneurysms, found a certain risk group consisting of patients aged 50 years or younger who had arterial hypertension and a history of smoking. Based on our case, we suggest that follow-up angiography should be considered in patients with known vascular anomalies, especially those with an APCA, which is intimately associated with saccular aneurysms.

References


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