De novo giant posterior cerebral artery aneurysm developing 25 years after basilar bifurcation aneurysm treatment using a Drake tourniquet: case report and implications for aneurysm follow-up

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The objective of this paper was to report a rare complication of basilar artery (BA) tourniquet treatment of a giant basilar tip aneurysm, and to discuss possible causes for the formation of a de novo giant posterior cerebral artery (PCA) aneurysm. A 34-year-old woman underwent satisfactory treatment of a ruptured giant basilar bifurcation aneurysm by BA ligation (Drake tourniquet) in 1985. She presented 25 years later with a new aneurysm in the left PCA, successfully treated by coil embolization. To the authors' knowledge, this is the first case of de novo aneurysm formation on a PCA, and the first de novo aneurysm reported as a complication of BA ligation therapy by Drake tourniquet. Long-term follow-up is necessary in patients with treated cerebral aneurysms, particularly those occurring in young patients, those with multiple aneurysms, those with complex posterior circulation aneurysms, and those undergoing flow diversion or flow-altering therapies.

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In 1979, Drake described proximal basilar artery (BA) ligation as a possible treatment for large bifurcation aneurysms that could not be directly clipped, particularly in giant, fusiform, or certain atherosclerotic aneurysms. This technique is only feasible if there is good collateral circulation from the posterior communicating arteries (PCoAs). Complications of this treatment include ischemic deterioration and partial thrombosis with risk of rupture; other long-term complications are rare.

The term “de novo” was first coined to describe the development of a new aneurysm in a previously angiographically normal site. Most of the previously described cases are in the anterior circulation. Location in the posterior circulation after previous aneurysm repair is very rare and the pathophysiology is poorly understood. De novo aneurysms can present with subarachnoid hemorrhage (SAH) several years after clipping of the original aneurysm. We report and discuss the first case of a de novo, unruptured giant aneurysm located in the posterior cerebral artery (PCA) after BA ligation using a Drake tourniquet.

Case Report

A 34-year-old woman presented in 1985 with an SAH due to a giant basilar bifurcation aneurysm (Fig. 1). Due to the large size and anticipated difficulties with direct clipping, this aneurysm was treated with BA occlusion by placement of a tourniquet below the superior cerebellar arteries (SCAs). Follow-up angiography showed a residual aneurysm remnant (Fig. 2), which filled from the anterior circulation via both PCoAs. The patient was symptom free postoperatively but presented 25 years later with headache and right-sided numbness. MRI with MR angiography showed a new, giant aneurysm arising from the left PCA at the P1/P2 junction with local vascular dysplasia (Fig. 3). The patient underwent initial aneurysm coil embolization.
in 2010 and repeat coil embolization a year later. Follow-up studies have shown a stable, small-neck remnant and no recurrence of the basilar bifurcation aneurysm (Fig. 4).

Discussion

Reports of de novo cerebral aneurysms are rare, with less than 20 described in the posterior circulation. Known risk factors include hypertension, young age, multiple aneurysms, smoking, Marfan syndrome, moyamoya disease, and female sex. The most probable mechanism is believed to be a change in the local hemodynamic environment. This change is believed to be responsible for the development of de novo aneurysm formation in the anterior circulation due to redirection of flow after carotid occlusion and may be responsible for aneurysm formation in patients with high-flow arteriovenous malformations. Hemodynamic changes in the cerebral circulation, however, are much more frequent than de novo aneurysms, so there must be other etiological factors involved. Remodeling of the aneurysm wall is a dynamic process in which genetic factors, inflammation, various proteins, vasospasm, and growth factors as well as hemodynamic factors play important roles.

Hunterian ligation by clip or tourniquet has been described previously for the treatment of unclippable basilar bifurcation aneurysms. A clip or ligature is used to occlude the BA in a perforator-free segment below the SCAs or between the SCAs and the basilar bifurcation. This is only possible when adequate collateral circulation is possible through the PCoAs and the advent of endovascular therapies with coils, stents, and other devices has rendered this procedure obsolete.

Most de novo posterior circulation aneurysms are located in the BA, presenting several years after clipping of an anterior circulation aneurysm. To our knowledge, this is the first reported case located in the PCA. Development at the P1/P2 junction fits the Rhoton criteria: location at the convexity of a curve, at the junction of 2 vessels, and following the direction of blood flow. The original basilar occlusion using a Drake tourniquet redirected blood flow from the BA/central bifurcation to a new posteriorly and laterally directed course from the PCoAs to the PCA at the P1/P2 junction. The fact that this aneurysm presented with mass effect rather than hemorrhage suggests that its pathophysiology was more likely influenced by hemodynamic change rather than factors that degrade the vessel wall, predisposing to rupture.

It is estimated that up to 2% of patients with ruptured cerebral aneurysms may harbor a second aneurysm, missed on initial imaging. The true incidence of de novo aneurysms may therefore be underestimated. In the present case, however, the initial and subsequent angiograms and multiple MR angiograms showed no evidence of a preexisting lesion at the P1/P2 junction.

The formation of de novo aneurysms has implications for screening; the incidence has been estimated at 0.3%—1.8%. The International Subarachnoid Aneurysm Trial (ISAT) study suggested an annual rupture rate from de novo and known additional aneurysms in patients with coiled aneurysms of 0.036%, while the rate in patients with previously clipped aneurysms is estimated at 0.06%. Although the Aneurysm Screening after Treatment for Ruptured Aneurysms (ASTRA) study showed that follow-up screening is not effective in patients with a history of SAH and aneurysm clipping, we believe that it
is still prudent to follow up patients at high risk for de novo aneurysms every 3–5 years, including young patients, those with Marfan syndrome, those with multiple or dysplastic aneurysms, and those with prior flow diversion or hemodynamic-altering aneurysm therapies.

This case report demonstrates the unique development of a de novo aneurysm in the PCA 25 years following BA ligation using a Drake tourniquet. The mechanism was likely due to the change in hemodynamics created by the original basilar occlusion. Although current endovascular therapies have rendered Hunterian ligation for basilar bifurcation aneurysms obsolete, the probable hemodynamic mechanism of aneurysm formation in this case has implications for the follow-up of patients with aneurysm treatment by flow-altering methods.

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