High-flow bypass and tandem microsurgical-endovascular occlusion of recurrent proximal middle cerebral artery aneurysms in a pediatric patient

Case report

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The authors report the case of a 7-year-old boy with headaches, in whom CT angiography showed multiple intracranial aneurysms from the terminus region of the right internal carotid artery through the proximal right middle cerebral artery (MCA). Initially, the patient underwent clip reconstruction of the M_1 segment. Multiple microsurgical and endovascular treatments were required because the largest of these aneurysms recurred several times over the next 1.5 years. The first recurrence was treated with stent coiling and the second by microsurgical occlusion of the MCA combined with the use of a radial artery graft in a common carotid artery–to-MCA bypass. The aneurysm again recurred and was treated by proximal coil occlusion 15 months after the first treatment session. At a 7.5-year follow-up examination, the aneurysms remained occluded. This case highlights the benefit of combined endovascular and microsurgical techniques in the treatment of a complex and unusual case of proximal MCA aneurysmal disease. The patient, now 15 years old, is neurologically intact and able to participate in all activities.
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KEY WORDS   •   stent   •   radial artery bypass graft   •   coil occlusion   •
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CEREBRAL aneurysms in the pediatric population commonly present with subarachnoid hemorrhage. Increased risk of aneurysmal rupture often calls for the treatment of unruptured aneurysms in this population, which otherwise has a long life expectancy. Such aneurysms are dynamic lesions; they tend to recur and new aneurysms tend to form.[3] Multiple intracranial aneurysms are common, especially in patients with connective tissue disorders. Thus, appropriate surveillance or detection of additional aneurysms is warranted. Middle cerebral artery aneurysms in children have been treated with a host of techniques, including clipping and/or extracranial-to-intracranial (EC-IC) bypass,[1,2,6] endovascular therapy,[7] trapping alone,[8] and bypass combined with endovascular therapy.[13] In children most MCA aneurysms are giant, fusiform, and difficult to treat with direct clipping alone.[1] They also can be calcified or dysplastic[13] and can involve proximal MCA perforators. Consequently, they are frequently treated by trapping (Hunterian ligation)[8] or flow reversal with potential EC-IC bypass in an attempt to preserve critical perforators.[13] Still, more creative and complex techniques may be required because flow reversal may fail if the patient is treated with bypass and proximal clip ligation because the aneurysm can continue to fill via the bypass.[13] We report an unusual case with long-term follow-up that highlights several difficulties in treating complex, dysplastic MCA aneurysms in a pediatric patient.

This article contains some figures that are displayed in color online but in black-and-white in the print edition.
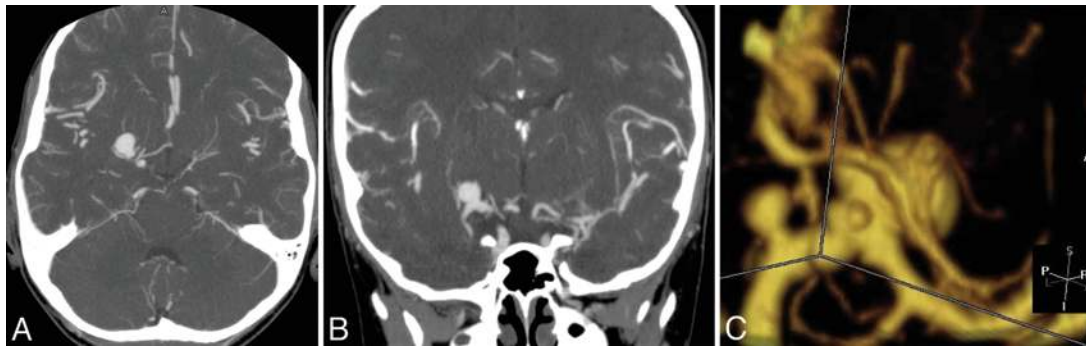
Fig. 1. Computed tomography angiograms of a 7-year-old boy with headaches show a large proximal right MCA aneurysm on both axial **(A)** and coronal **(B)** views. The neck of the aneurysm is near the exit of the anterior temporal branch of the MCA. A reconstructed 3D image shows multiple small aneurysms of the proximal right MCA **(C)**.

Case Report

History and Presentation. This 7-year-old boy with headaches underwent CT angiography, which showed multiple intracranial aneurysms from the terminus region of the right ICA through the proximal right MCA (Fig. 1). The patient's family was counseled about the risks and benefits of intervention; they opted for microsurgical treatment of the intracranial aneurysms.

Initial Operation. At surgery, 7 aneurysms of the ICA terminus and proximal MCA were found. Several appeared to have thrombosed, and others resembled small blister-type aneurysms. The largest lesion involved the MCA just proximal to the takeoff of the anterior temporal artery. Multiple clips were used to reconstruct the proximal MCA and to obliterate the aneurysms. Intraoperative angiography demonstrated patency of the MCA on the right side with well-occluded aneurysms (Fig. 2A and B).

The patient remained neurologically intact in the ICU. However, on the 1st postoperative day, postoperative angiography showed thrombosis of the right MCA, and CT perfusion imaging showed mildly asymmetrical perfusion of the right MCA territory (Fig. 2C). The boy was started on Aggrastat (tirofiban, Iroko Cardio International) and monitored closely in the ICU. On postoperative Days 2 and 3, angiography showed serial improvement in MCA flow and the patient was switched from Aggrastat to aspirin. He was extubated after the final angiogram and transferred from the ICU on postoperative Day 6. He was discharged home on postoperative Day 7 without neurological deficit.

Second Operation. An angiogram 5 months later demonstrated a complex recurrence (Fig. 3A and B) at the site of the large proximal MCA aneurysm. At that point, the recurrence was treated with endovascular techniques. First, a 4 × 15–mm Neuroform stent (SMART Therapeutics, Inc.) was placed across the neck of the aneurysm. Thirteen coils were inserted into the aneurysm and more than 90% occlusion was achieved (Fig. 3C). The patient made an excellent recovery.

Third Operation. At his 1-year follow-up examination, the now 8-year-old boy was found to have a substantial recurrence, including a large 8-mm aneurysm and two smaller 2-mm aneurysms (Fig. 4 left). At this point, no further endovascular treatment was attempted. Instead, the patient underwent a right common carotid artery–to-MCA bypass with a radial artery interposition graft. The bypass was anastomosed to an M_2 recipient vessel, and the MCA (M_1) was clip ligated just distal to the aneurysm (Fig. 4 right) in an effort to preserve vital perforators arising from the proximal MCA. The patient tolerated this procedure well and was discharged after a
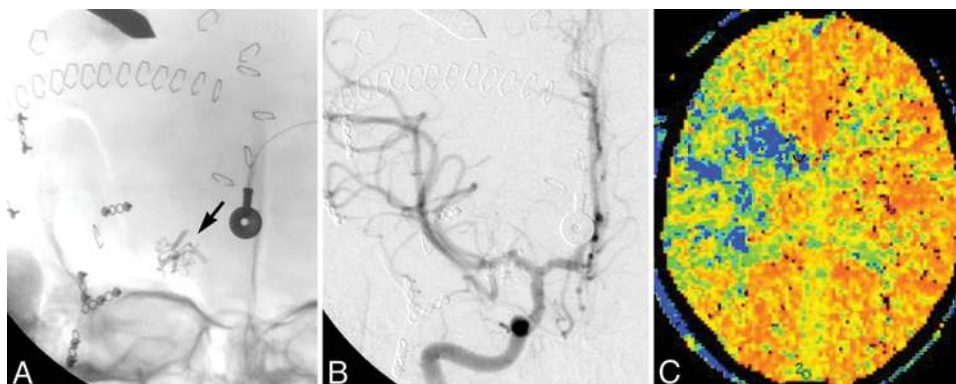

Fig. 2. Intraoperative anteroposterior unsubtracted **(A)** and digitally subtracted **(B)** angiograms demonstrate excellent reconstruction of the MCA with multiple clips (A, *arrow*) and patency of the distal MCA **(B)**. Postoperative CT perfusion scan shows asymmetry in perfusion with increased mean transit time in the right hemisphere on axial imaging **(C)**.

FIG. 3. Anteroposterior angiogram **(A)** obtained 5 months later demonstrated a recurrence of the largest aneurysm and continued aneurysmal disease of the proximal MCA on a 3D reconstruction **(B)**. Both the exit of the anterior temporal artery and the MCA bifurcation can be seen distal to the largest aneurysm. The recurrence was treated by placing a Neuroform stent across the aneurysm neck and primary coiling. The anteroposterior angiogram after both stenting and coiling is shown **(C)**.

5-day hospital stay. His only neurological deficit was a subtle left upper-extremity pronator drift that developed on postoperative Day 2.

Fourth Operation. Three months later, the patient returned for follow-up. The largest aneurysm had failed to thrombose with distal clip ligation. Importantly, this angiogram also showed a lack of perforators arising from the origin of the aneurysm (Fig. 5A). The boy therefore underwent coil embolization of this recurrent M_1 aneurysm, including proximal M_1 occlusion (Fig. 5B and C).

Postoperative Course. One year after the previous coiling (now 27 months since the initial therapy), a common carotid artery injection angiogram showed no further recurrence and a patent bypass (Fig. 6A). Angiography obtained at 4 years (Fig. 6B) and at 7 years (Fig. 6C) also confirmed a patent bypass and no further growth of the aneurysm. At his last follow-up, the now 15-year-old
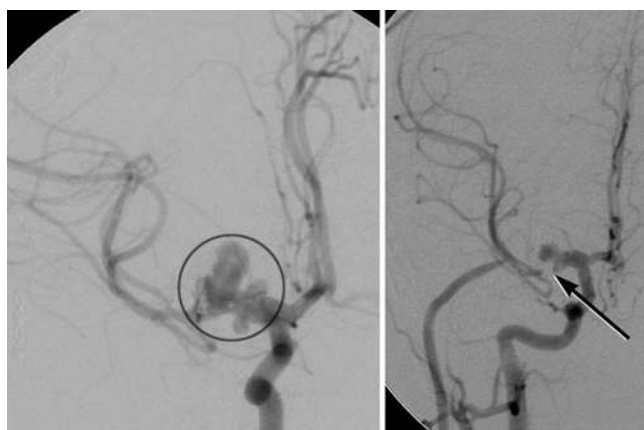

FIG. 4. Anteroposterior angiograms obtained 6 months later (1 year after initial treatment) showed a significant recurrence of the large MCA aneurysm (**left**, *circle*) as well as smaller recurrences nearby. A common carotid artery–MCA (M_2) bypass was performed using a radial artery graft. **Right:** Postoperative anteroposterior angiogram demonstrated both the distal end of the bypass and clip ligation distal to the bypass *(arrow)*.
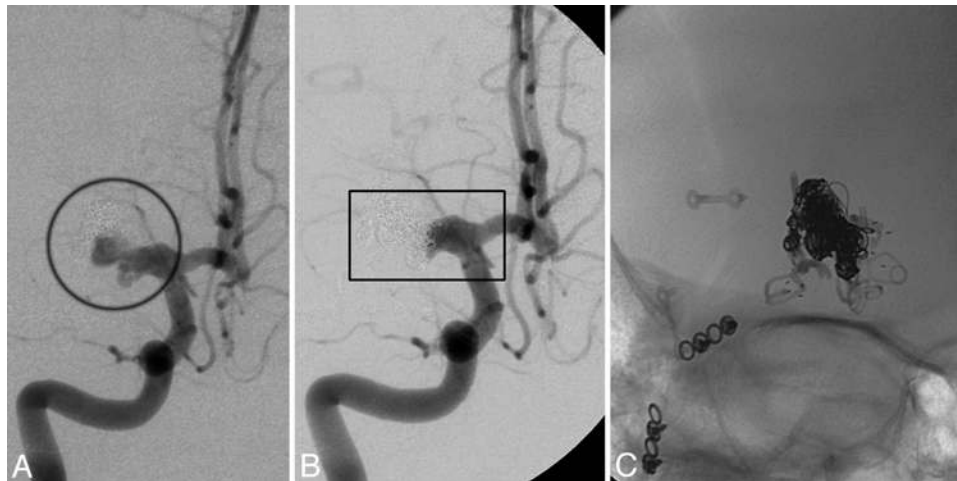
patient remains neurologically intact and actively participates in high school sports.

Discussion

Pediatric patients harbor between 0.5% and 4.6% of all aneurysms, which usually occur in boys after the age of 10 years.[12] Several causes have been implicated, including genetic connective tissue disorders, infection, and both blunt or birth trauma.[12] Dissecting aneurysms in children are the most common type and can cause infarction, hemorrhage, or mass effect.[5] Spontaneous dissecting aneurysms of the MCA in children often cause ischemia, but these lesions also can rupture.[8] In a multicenter report of 22 pediatric aneurysms, Proust et al.[9] noted that the locations of pediatric intracranial aneurysms differed from the most common sites in adults: 36.4% of pediatric aneurysms were located at the ICA bifurcation, a relatively rare site for aneurysms in adults. The MCA was involved by 36.4% of the pediatric aneurysms, half of which involved the distal MCA. Giant aneurysms constituted 14% of all aneurysms. In their series, all but one patient became symptomatic with hemorrhage. In patients with subarachnoid hemorrhage, dissecting aneurysms are thought to be associated with a poor prognosis, and rehemorrhage from these aneurysms is associated with a high rate of mortality.[12]

Our patient did not present with subarachnoid hemorrhage; however, the morphology and number of aneurysms were ominous. Because of the otherwise long life expectancy of a 7-year-old patient, definitive treatment was indicated. This case is unusual for several reasons. First, these MCA aneurysms arose between the ICA bifurcation and the anterior temporal branch, an uncommon location for intracranial aneurysms. The number and morphology of the aneurysms (Fig. 3B) were impressive as was the continued recurrence of the aneurysms. The largest aneurysm had both fusiform and saccular components while the smaller ones consisted of aneurysms that were either thrombosed on surgical exploration or

Fig. 5. An anteroposterior angiogram obtained 3 months after the third operation (**A**, ICA injection) showed that the residual aneurysm *(circle)* had not thrombosed with clip ligation of the MCA distal to the aneurysm. The patient therefore underwent further coiling (**B**, *rectangle*) of the aneurysm and parent vessel, which demonstrated a good result on the final subtracted (**B**) and unsubtracted images (**C**).

that filled robustly and appeared blister-like. Connective tissue disorders such as fibromuscular dysplasia, Ehlers-Danlos Type IV, and Marfan syndrome have been implicated in similar cases.[4,11]

The recurrences after each treatment session also suggest the possibility of a disorder of the vascular connective tissue. However, no connective tissue disorder has been diagnosed in our patient; neither is there a familial history of sudden death related to subarachnoid hemorrhage nor a family history of aneurysms. Neither clip reconstruction nor stent-assisted coil embolization was enough to obliterate the patient's aneurysms permanently. This finding has also been reported in pediatric intracranial aneurysm series.[3,10] In a study of 32 children with 43 aneurysms, the recurrence rates in both surgically treated (6%) and endovascularly treated (18%) patients were high with 19% of those in the endovascular group also demonstrating de novo aneurysm formation.[10]

Although our patient did well, the finding of CT perfusion asymmetry after initial clip reconstruction underlines the need for careful observation to ensure adequate perfusion of the treated hemisphere when patients with MCA aneurysmal disease are treated with coiling, clipping, stenting, trapping, or bypass. Furthermore, placement of flow-diverting devices in the MCA is associated with a similar risk of MCA territory ischemia, especially for MCA perforators that may be jailed by the low-porosity device.

Fig. 6. Common carotid artery injection anteroposterior angiogram 1 year later (2.25 years since the first treatment) confirmed patency of the distal (**A**) portion of the radial artery graft with a stable M_1 stump and no further aneurysm growth. The 4-year follow-up anteroposterior angiogram showed continued patency of the bypass on common carotid artery injection (**B**). The 7.5-year follow-up angiogram (**C**) again confirmed patency of the radial artery graft with a stable M_1 stump and no further aneurysm growth.

Conclusions

This case highlights the unusual vascular aneurysmal pathology that can occur in the pediatric population as well as the novel combined surgical and endovascular techniques that can be used in multimodal treatment. The potential implication of connective tissue disorder in such pediatric patients is reflected by both the short time to recurrence and the size of the recurrent aneurysms. In children especially, the dynamic nature of both recurrences and de novo aneurysm formation requires continued follow-up in pediatric patients without signs or symptoms. The goal is to arrest the progression of the disease. Children also tend to have unusual aneurysms in less common locations than adults. The resilience of children in being able to undergo multiple large, complex operations is also demonstrated by this patient who remains neurologically intact and has done remarkably well during the 7.5 years since his first operation.

Disclosure

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