Stent-assisted coil embolization of a symptomatic middle cerebral artery aneurysm in an infant

Case report


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Pediatric intracranial aneurysms are rare and challenging to treat. Achieving efficacy and durability of aneurysmal occlusion while maintaining parent vessel patency requires innovative treatment strategies, especially in cases in which aneurysmal location or morphology pose substantial morbidity associated with microsurgical treatment. In the last 3 decades, endovascular treatments have had a remarkable evolution and are currently considered safe and effective therapeutic options for cerebral aneurysms. While endovascular techniques are well described in the English literature, the endovascular management of pediatric aneurysms continues to pose a challenge. In this report, the authors describe the case of a 9-month-old infant who presented with a 1-day history of acute-onset left-sided hemiparesis and left facial droop. Imaging revealed a large symptomatic saccular middle cerebral artery aneurysm. Treatment included successful stent-assisted aneurysm coiling. At follow-up, the patient continued to fare well and MR angiography confirmed complete occlusion of the aneurysm dome. This case features the youngest patient in the English literature to harbor an intracranial aneurysm successfully treated with stent-assisted coiling. Based on this experience, endovascular intervention with vascular reconstruction can be safe and effective for the treatment of infants and could further improve prognosis; however, further studies are necessary to confirm these findings. (http://thejns.org/doi/abs/10.3171/2014.7.PEDS1449)

Key Words • embolization • pediatric intracranial aneurysm • stent-assisted coiling • vascular disorders

Intracranial aneurysms in the pediatric population occur infrequently (prevalence 0.5%–4.6%) and are challenging to treat.5,6,9,22,25,26,30 Endovascular treatment of ruptured and unruptured cerebral aneurysms in children has been reported with increasing frequency.1,4,7,14,19,28,29,36 This represents a change in treatment paradigm, as microsurgical techniques were previously used exclusively.33 Coiling of wide-necked aneurysms continues to pose a significant challenge, as parent vessel patency is at risk. Numerous innovative devices and therapeutic strategies have been developed to render the endovascular treatment of wide-necked aneurysms more effective and durable, including stent-assisted coiling (SAC), balloon-assisted coiling, liquid embolization, and flow diversion with the Pipeline embolization device (Cook). These techniques have been used in adults but have been rarely reported in the infant population. To our knowledge, the following case involves the youngest patient to undergo successful SAC, as reported in the English-language literature.

Case Report

History and Examination. A previously healthy 9-month-old male infant presented to the emergency department with a 1-day history of the acute onset of left-sided hemiparesis and left facial droop. Nondenhanced CT scanning of the head showed a round hyperdense lesion measuring 1 cm in its greatest diameter, located within the right sylvian cistern along the course of the right middle cerebral artery (MCA); however, there was no evidence of subarachnoid hemorrhage (Fig. 1A). Magnetic resonance imaging of the brain revealed an acute infarction of the right corpus striatum and anterior limb of the internal capsule with rostral extension into the corona radiata (Fig. 1B). Magnetic resonance angiography (MRA) of the head displayed an 11-mm saccular aneurysm with a wide...
Stent-assisted coil embolization of an MCA aneurysm in an infant

Fig. 1. A: Noncontrast axial head CT scan shows a hyperdense 1-cm lesion within the right sylvian cistern along the course of the right MCA (black arrow). B: Diffusion-weighted axial MR image demonstrates abnormal impeded diffusion in the right corpus striatum and anterior limb of the right internal capsule with rostral extension into the corona radiata. C: Axial MR angiogram reveals a saccular 9 × 9 × 10–mm right proximal MCA aneurysm with a wide neck close to the internal carotid artery terminus, just distal to the takeoff of the right posterior communicating artery. D: Axial 3D MR angiogram shows patent proximal and distal right MCA segments but diminished flow contrast opacification as compared with that in the left MCA, suggestive of relatively slow flow. No additional aneurysms, dissections, or intrinsic stenosis are demonstrated.

The patient received dual antiplatelet medications (clopidogrel bisulfate 4 mg and aspirin 40 mg) prior to the procedure. After inducing general anesthesia via endotracheal tube and utilizing a routine sterile technique, we introduced a 4-Fr Pinnacle sheath (Terumo) into the left common femoral artery with the aid of ultrasound guidance. A 4-Fr Glidecath (Terumo) was advanced to the horizontal segment of the right petrous internal carotid artery. Under roadmap guidance, the right M1 segment was selectively catheterized using an XT microcatheter (Stryker), and a 3 × 20–mm Neuroform EZ stent (Stryker) was deployed across the aneurysm neck (Fig. 2A and B). An SL-10 microcatheter (Boston Scientific) was advanced through the stent into the aneurysm, which was then obliterated with Target detachable coils (Stryker). Postembolization runs revealed thrombus formation at the distal portion of the stent (Fig. 2C). The SL-10 microcatheter was removed from the aneurysm and placed within the thrombus, allowing the administration of 2 mg of abciximab (Eli Lilly) within the thrombus, leading to complete patency of the vessel (Fig. 2D). There was no evidence of a delay in transit time, and distal vessels were patent.

Posttreatment Course. The patient was admitted to the neonatal intensive care unit and maintained on 4 mg of clopidogrel bisulfate and 40 mg of aspirin daily. Following the procedure, the patient remained at baseline with left-sided hemiparesis. Cerebral angiography 24 hours postembolization revealed complete obliteration of the aneurysm dome with patency of M1 and partial resolution of the preprocedural M1 stenosis (Fig. 3). The patient was discharged to home on Day 7 with symmetric facial movements and an improving left-sided hemiparesis. Mild language delay was detected at an age of 21 months; however, the patient continues to achieve appropriate milestones as he grows into childhood. Clopidogrel bisulfate was discontinued 6 months following the procedure, while aspirin has been continued. Follow-up MRI and/or MRA were performed at 2 months and revealed chronic ischemic changes within the right basal ganglia and internal capsule. The aneurysm remained obliterated, while the MCA was patent with no evidence of stenosis. Repeated MRI and/or MRA at 6, 12, 18, and 24 months postembolization displayed stable aneurysmal healing with minimal stable filling at the aneurysm neck (Fig. 4).

Discussion

Pediatric cerebral aneurysms are rare and account for less than 5% of all intracranial aneurysms.6,9,22,25,26,30 Such aneurysms tend to be larger, more complex in angioarchitecture, and have a higher incidence in boys, as compared with cerebral aneurysms in the adult population. Seventy-six percent of pediatric aneurysms are located in the anterior circulation, with MCA aneurysms three times more prevalent than other anterior circulation locations.9 In addition, patients presenting with hemorrhage are younger (mean age 4.3 vs 6.7 months, respectively) and tend to have smaller aneurysms than those presenting with nonhemorrhagic symptoms. Our case—a 9-month-old male infant harboring a large MCA aneurysm who presented with an acute neurological deficit in the setting of an ischemic stroke—is consistent with the above characteristics. In addition, 21% of pediatric aneurysms can be associated with trauma or connective tissue disorders (polycystic kidney disease, collagen vascular disease, or aortic coarctation); however, this was not the case in our patient.

The most appropriate treatment for pediatric cerebral
aneurysms is currently under debate, and updated guidelines have yet to be defined. Contemporary endovascular and microsurgical techniques are considered equivalent in effectiveness and safety when used in appropriate pediatric cases. Extrapolating from adult trials, such as the Barrow Ruptured Aneurysm Trial (BRAT) and the International Subarachnoid Aneurysm Trial (ISAT), it is plausible that endovascular methods will provide better outcomes than microsurgical treatment. In addition, there is minimal tolerance for blood loss in younger patients, thus making microsurgery much more challenging.

The existing literature indicates that microsurgical therapy leads to higher rates of complete aneurysm obliteration (92%–93% vs 79%–82% for endovascular treatment) and lower rates of recurrence (0% vs 14% for endovascular treatment) in children. Despite the reported superiority of the open surgical approach as regards obliteration and recurrence rate, the last few decades have witnessed a change of paradigm characterized by a gradual shift from traditional microsurgical approaches toward endovascular treatment of aneurysms in adults and pediatric patients. The ongoing shift was strengthened in 2002 with the publication of the ISAT, which reported a statistically significant 7.4% absolute 1-year disability-free survival advantage favoring coiling. Unfortunately, wide-necked (neck > 4 mm or aspect ratio < 2) and fusiform aneurysms were typically not included in that study; thus, the standard of care for these aneurysms is still under debate.

As mentioned earlier, wide-necked aneurysms remain a challenge for neurosurgeons and interventional radiologists.

Fig. 2. Digital subtraction angiograms, anteroposterior view, of an 11×6-mm wide-necked M1 aneurysm with mild kink/stenosis (A) involving the distal aspect of the parent vessel next to the aneurysm (arrow). A 3×20-mm Neuroform EZ stent was deployed across the aneurysm neck (arrow, B). Multiple coils were inserted under stent protection (C), and postembolization runs revealed thrombus formation at the distal aspect of the aneurysm neck within the parent vessel. Arrows indicate both ends of the stent. Bracket indicates the intraluminal thrombus. Postchemolytic runs revealed patency of the right M1 with minimal stenosis within the segment that was present preoperatively (D). There was no evidence of a delay in transit time. Completion angiography shows occlusion of the aneurysm with a residual neck.

Fig. 3. Digital subtraction angiogram obtained 24 hours after SAC embolization is significant for patency of the right M1 with no evidence of stenosis or thrombosis and complete obliteration of the aneurysm dome.

Fig. 4. Coronal contrast-enhanced MR angiogram obtained 24 months after SAC embolization shows stable aneurysmal healing (arrow), with stable filling at the aneurysm neck and patent proximal and distal right MCA segments with flow contrast opacification equal to that in the left MCA.
In the pediatric population, direct aneurysm clipping has proved to be the most definitive and durable treatment for such lesions, but it is often hazardous and at times impossible to perform because of difficult surgical access, the incorporation of a parent vessel and/or perforators, and requisite proximal arterial ligation or trapping. Since the introduction of first-generation neurovascular stents into the endovascular surgery armamentarium in the late 1990s, SAC embolization has become a safe and effective option in adults to treat intracranial wide-necked aneurysms. Initially designed to act as a scaffold to prevent intrasaccular coil protrusion into the parent artery lumen, these stents have been shown to divert blood from the aneurysm inflow zone, to promote intraaneurysmal stasis and thrombosis and thereby prevent or reduce coil compaction by changing intraaneurysmal flow, and to provide luminal matrix for orifice endothelialization.

The art of embolization requires a delicate balance between the need to pack a fragile aneurysm optimally to prevent hemorrhage and the risk of intra procedural aneurysm rupture and coil herniation. Often, a residual aneurysm neck or fundus is left behind on the immediate postcoiling angiogram. In our patient, we purposely kept the base of the aneurysm patent, as the aneurysm neck harbored lenticulostriate perforating vessels. Follow-up MRA at 2, 6, 12, 18, and 24 months postembolization revealed complete occlusion of the dome with stable aneurysm neck filling and partial normalization of the carotid artery lumen. Contrast-enhanced MRA (CE-MRA) showed patent proximal and distal right MCA segments and demonstrated flow contrast opacification equal to that of the left MCA (Fig. 4). In addition, no evidence of ischemia was observed on follow-up MRI, correlating with the nonfocal neurological examination. Our group favors CE-MRA over time-of-flight MRA for the follow-up of coiled aneurysms, given the significant artifacts related to the latter technique. Time-of-flight MRA is based on the direction of protons flowing into the imaging voxel. Flow-related artifacts can occur if nonlaminar flow is present. These artifacts can be exaggerated from the metal artifact related to the coil mass within the aneurysm. Contrast-enhanced MRA is similar to digital subtraction angiography (DSA) since they both measure opacification of the vessel lumen.

The significance of a residual nonprogressive neck is uncertain and the center of current debate. Studies in adult populations have shown that angiographic recanalization of a coiled aneurysm has a rerupture rate of 0.57%–1.1%, and reintervention appears to be safe. Despite the low annual rerupture rates, the long-term cumulative risk is not negligible in children; therefore, the necessity of lifelong angiographic follow-up must be considered in the treatment algorithm based on the remaining life expectancy in the pediatric population. Our experience suggests that endovascular intervention techniques with vascular reconstruction are potentially safe and effective in infants and could further improve prognosis. However, further studies are required to confirm our findings.

Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Pandey. Acquisition of data: Pandey, Chaudhary, Gemmete, Maher. Analysis and interpretation of data: Savastano, Chaudhary, Gemmete, Garton. Drafting the article: Savastano. Critically revising the article: Savastano. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Pandey.

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References


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