

## Extracranial-intracranial bypass in a neonate with multiple ruptured middle cerebral artery aneurysms: illustrative case

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**BACKGROUND** Reports of ruptured neonatal aneurysms are rare in neurosurgical literature. Pediatric aneurysms differ from adult aneurysms, notably in morphology, size, number, and risk of rerupture. Many authors report experience with clipping, citing durability and decreased use of radiation as benefits over endovascular intervention. Few authors report extracranial-to-intracranial bypass because small pediatric vessels make this option challenging. The authors discussed a case of a newborn with multiple ruptured aneurysms, one of the youngest reported cases involving extracranial-intracranial bypass.

**OBSERVATIONS** A 3-week-old baby presented with hemorrhage from multiple complex middle cerebral artery (MCA) aneurysms. Because of young age, endovascular intervention was not possible; therefore, the patient received craniotomy. Upon exploration, clip reconstruction was impossible; the vessel was trapped, and superficial temporal artery (STA)-MCA bypass was performed. The recipient vessel diameter was 0.3 mm. The postoperative course was complicated by seizures as well as symptomatic vasospasm, which was treated with intraarterial verapamil and ventriculostomy. At last follow-up, the patient was developing normally and was ambulatory with minimal deficit.

**LESSONS** This case, one of the youngest patients reported, highlighted details of pediatric aneurysm management, such as propensity for multiple/fusiform aneurysms and high risk of re-hemorrhage, with significant mortality. The authors recommended aggressive, early intervention in pediatric aneurysms at centers with surgeons familiar with both endovascular intervention and cerebral bypass.

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**KEYWORDS** pediatric; aneurysm; subarachnoid hemorrhage; extracranial-intracranial bypass; clipping

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### Illustrative Case

#### Clinical Presentation and Imaging

Our patient presented at 3 weeks of age to the emergency department for sudden crying and apnea. Examination demonstrated a lethargic, irritable infant with agonal respirations. The fontanel was bulging. Pupils were 4 mm and reactive; grimace was symmetric. All extremities moved spontaneously. The patient was born at 39 weeks' gestation via Cesarean section. She has one healthy 3-year-old brother. Her great-grandmother died at age 75 from a ruptured cerebral aneurysm.

Computed tomography (CT) demonstrated subarachnoid and intraventricular blood with hydrocephalus. CT angiography (CTA) showed a 5.3-mm blister aneurysm incorporating the M1 segment

**ABBREVIATIONS** CT = computed tomography; CTA = CT angiography; LUE = left upper extremity; MCA = middle cerebral artery; STA = superficial temporal artery.

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**FIG. 1.** Preoperative CTA three-dimensional reconstruction demonstrating multiple right MCA aneurysms.

of the right middle cerebral artery and a second saccular aneurysm arising from the proximal right M1 segment (Fig. 1).

### Operative Intervention

Endovascular intervention was not possible, so a pterional craniotomy was performed. The right STA was preserved, and the bone flap was removed in multiple pieces because of the immature skull. Upon dural opening, intracranial pressure was extremely high and remained elevated despite mannitol, head elevation, hyperventilation, and ventricular puncture. Ultimately, the lamina terminalis was fenestrated via microscopic dissection, allowing enough relaxation to continue exploration.

New hematoma was found in the temporal lobe, indicating re-hemorrhage since admission scans. The hematoma was evacuated. The distal aneurysm was much larger than imaging indicated, with green discoloration, suggesting pseudoaneurysm or mycotic aneurysm. Almost the entire M1 segment was diseased, including the anterior temporal artery takeoff. The smaller proximal aneurysm arose from a lenticulostriate artery. Clip reconstruction of the vessel was impossible without leaving the patient with substantial risk of future hemorrhage; therefore, we proceeded with vessel trapping and bypass.

The parietal STA was harvested. The vessel diameter was 0.3 mm, requiring fishmouthing to create the appropriate diameter for anastomosis. The recipient M2 was cross-clamped, and the artery was incised and irrigated with heparin. Anastomosis was performed with 10-0 nylon suture at each at heel and toe, the clips were released, and flow through the anastomosis was confirmed.

The proximal M1 segment was identified, and a clip was placed obliquely such that the vessel and proximal aneurysm were occluded but the lenticulostriate vessel remained patent. The distal diseased M1 segment was identified and similarly clipped to occlude the vessel and preserve the distal lenticulostriate. These clips completely trapped the diseased M1 (Figs. 2–4). Blood pressure was then augmented 20% above normal. Upon closure, the brain remained edematous, so the



**FIG. 2.** Postoperative CTA three-dimensional reconstruction after trapping and bypass, demonstrating complete occlusion of both the proximal lenticulostriate aneurysm and the distal diseased segment, with preserved flow in the distal MCA from STA-MCA bypass.

bone flap was loosely reapproximated using suture, allowing the bone to mobilize outward with further swelling, alleviating intracranial pressure and maintaining graft patency.

Postoperatively, the patient was placed on aspirin 20 mg daily. The patient moved all extremities, with mild left upper extremity (LUE) paresis. She developed seizures on postoperative day 1 and symptomatic vasospasm on postbleed day 9, which required intraarterial verapamil and a ventricular drain, which were later weaned and removed.

At 16 months of age, the patient was developmentally normal and ambulatory, with only mild LUE spasticity. She remains on levetiracetam for breakthrough seizures. CTA demonstrated patency of the bypass (Fig. 3).

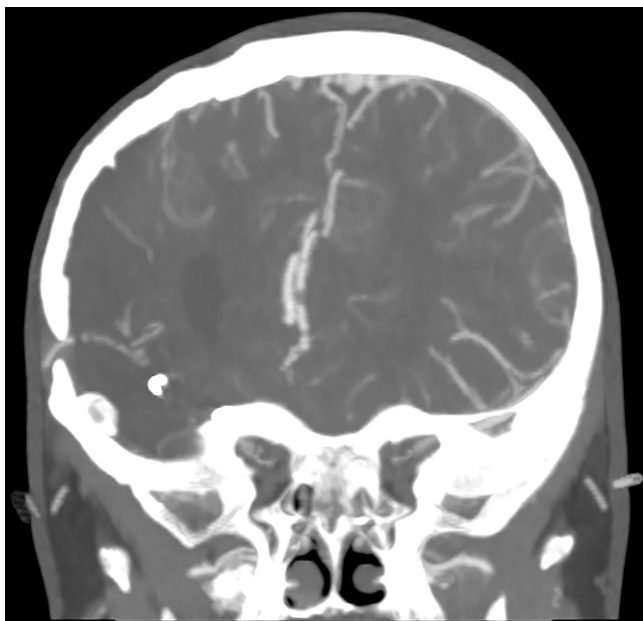
## Discussion

### Observations

Pediatric aneurysms are unusual and rare enough that large autopsy studies have found no incidental aneurysms in pediatric populations below certain ages.<sup>1</sup> Studies examining neonatal patients are rarer and restricted to case reports or small series. Therefore, data are limited and conflicting.

Pediatric aneurysm etiology is not clear because these aneurysms lack many risk factors that contribute to adult aneurysms, such as tobacco use. Some factors remain relevant, such as genetics and illnesses such as polycystic kidney disease and aortic coarctation.<sup>2</sup> In our patient's case, no abnormality was found.

Pediatric aneurysms also differ in size, location, morphology, and prognosis. Studies document a large proportion of aneurysms (sometimes nearly 50%) occurring in the MCA distribution, and up to 25% to 33% may occur in the posterior circulation.<sup>3–5</sup> This finding contrasts with the often quoted "Acomm" and "Pcomm" in adults. Pediatric populations also have higher rates of giant aneurysms, possibly up to 30%,<sup>3</sup>



**FIG. 3.** Coronal CTA demonstrating patency of the STA-MCA bypass at 16-month follow-up.

and higher rates of blister morphologies, possibly as high as 70%, particularly in MCA aneurysms.<sup>6</sup>

These details impact diagnosis and treatment because pediatric aneurysms are rarely found incidentally. A French series of 43 pediatric aneurysms found that >80% presented with hemorrhage.<sup>7</sup> Another series of 28 patients reported hemorrhage in 67%, infarct in 7%, and cranial nerve palsies in 4%.<sup>6</sup> Only one hemorrhage presented incidentally, during an ultrasound for sickle cell anemia. Of patients who experienced

hemorrhage, the re-hemorrhage rate was 10% and was universally fatal. This high re-hemorrhage rate is preserved across multiple studies, with another study reporting a rate of 52%.<sup>8</sup> Re-hemorrhage resulted in high morbidity rates and, in many cases, was the cause of death.<sup>6,8</sup>

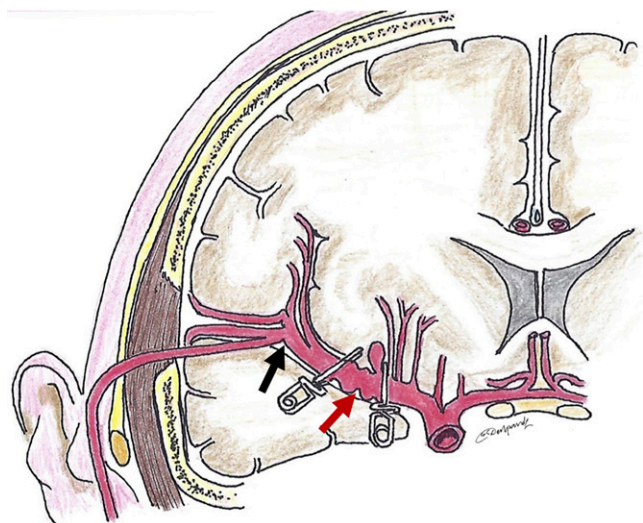
The high re-hemorrhage rate and morbidity are important because pediatric patients otherwise tolerate aneurysmal rupture relatively well. Many studies document positive outcomes. One study of 22 patients documented favorable outcomes in 14 (63.6%) patients and unfavorable outcomes in 8 (36.4%) patients. Unfavorable outcomes were attributed to the initial hemorrhage, rebleeding, and surgical complications, including an intraoperative rupture of an MCA aneurysm requiring vessel ligation.<sup>8</sup> Another study reported positive outcomes, with 63.4% of 43 patients cured without any residual symptoms, 19.5% with loss of academic time but normal life, 4.8% with severe disability, and a mortality rate of 12.3%.<sup>7</sup>

### Lessons

Considering the likelihood of a good outcome and the risk/morbidity of re-hemorrhage, we recommend aggressive, early intervention. Given the high rate of blister aneurysms and frequent MCA location, we recommend that centers treating these pathologies have both skilled open and endovascular teams. Many authors report good outcomes with clipping or bypass,<sup>9,10</sup> and more recently there have been reports of successful endovascular intervention in neonatal patients. One author reported a successful coiling of an M2 aneurysm without recurrence in 10 months of follow-up.<sup>5</sup> However, in our patient, given the tiny vessel caliber, dissecting morphology, and surgical team's familiarity with bypass, we opted to forgo an attempt at endovascular intervention.

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**FIG. 4.** Artist's rendering of the clip positioning. The proximal clip is positioned to occlude the aneurysm while maintaining patency of the lenticulostriate vessel associated with it, whereas the distal clip is placed to completely eliminate the diseased MCA segment. Red arrow points to the blister MCA aneurysm. Black arrow points to the STA-MCA anastomosis on the M2 segment.

**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**

Conception and design: Kahn. Acquisition of data: Kahn, Russell, Lin, Klopfenstein. Analysis and interpretation of data: Kahn, Russell, Klopfenstein. Drafting the article: Kahn, Deshpande, Russell. Critically revising the article:

Kahn, Deshpande, Russell, Klopfenstein. Reviewed submitted version of manuscript: Kahn, Deshpande, Klopfenstein. Approved the final version of the manuscript on behalf of all authors: Kahn. Administrative/technical/material support: Lin. Study supervision: Klopfenstein.

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