Infantile traumatic pericallosal aneurysm: illustrative case

Zachary S. Hubbard, MD, MBA, Conor M. Cunningham, MD, Brian F. Saway, MD, Matthew J. Triano, MD, Aaron T. Miller, DO, Guilherme Porto, MD, Libby Kosnik Infinger, MD, MPH, and Alejandro M. Spiotta, MD

Department of Neurosurgery, Medical University of South Carolina, Charleston, South Carolina

BACKGROUND Traumatic aneurysms are a rare sequela of nonaccidental head trauma in infants. The rate of nonaccidental trauma (NAT) in the pediatric population is increasing; therefore, traumatic aneurysms are an important consideration in the evaluation of pediatric patients with abusive head trauma.

OBSERVATIONS A 24-day-old infant with no significant past medical or birth history presented with twitching and poor oral intake for 1 day. The patient was found to have bilateral subdural hematomas, multifocal contusions, and traumatic subarachnoid hemorrhage. NAT work-up was remarkable for a period of repeated and prolonged abuse. Magnetic resonance angiography revealed a right pericallosal traumatic aneurysm that was treated by means of coil and Onyx embolization.

LESSONS Traumatic intracranial aneurysms are a rare but serious sequela of pediatric abusive trauma. Traumatic intracranial aneurysms should be considered in the setting of intracranial pathology associated with high-energy trauma. Despite new methods for the management of traumatic aneurysms, this pathology remains challenging to identify and treat, and the prognosis remains poor because of the diffuse injury often involved in these patients.

https://thejns.org/doi/abs/10.3171/CASE23600

KEYWORDS aneurysm; traumatic cerebral aneurysm; pediatrics; cerebrovascular trauma; neurotrauma

Nonaccidental trauma (NAT) is a leading cause of infantile injury and death in the United States. An estimated 2.07 children per 100,000 died in the United States as a result of abuse or neglect in 2010. Over 50% of all fatalities resulting from child abuse occur in those under the age of 12 months. Abusive head trauma is most commonly characterized by skull fracture, subdural hematoma (SDH), cerebral edema, and retinal hemorrhages. Traumatic cerebral aneurysms (TCAs) are a rare sequela of abusive head trauma in infants. The rate of NAT in the pediatric population is increasing; therefore, TCAs are an important consideration in the evaluation of pediatric patients with abusive head trauma. We report a case of a 24-day-old infant who had a TCA arising from the right pericallosal artery.

Illustrative Case

A 24-day-old infant with no significant past medical or birth history presented with a 1-day history of twitching and poor oral intake.

Neurological examination revealed increased tone throughout all four extremities. The child was found to have retinal hemorrhages on fundoscopic examination. Due to ongoing seizures, the patient was intubated for seizure control and treated with levetiracetam, phenobarbital, and midazolam infusion. Computed tomography (CT) of the head revealed bilateral acute-on-chronic SDHs, multifocal cortical hypodensities concerning for stroke, and encephalomalacia bilaterally (Fig. 1). Magnetic resonance imaging (MRI) of the head revealed bilateral acute-on-chronic SDHs, multifocal intraparenchymal hematomas, and subacute subarachnoid hemorrhage (Fig. 2). Magnetic resonance angiography (MRA) revealed a 3-mm aneurysm arising from the right pericallosal artery (Fig. 3). The clinical picture was consistent with NAT, and the case was referred to law enforcement and the Department of Social Services.

Given the findings on MRA, diagnostic cerebral angiography was performed (Fig. 4A and B). Performing diagnostic angiography and...
administering subsequent treatment in infants present specific challenges. A small patient size portends risks with radiation, contrast exposure, vascular access, and vessel navigation. Our institution has a standardized protocol for the performance of infantile diagnostic cerebral angiography addressing these concerns. We perform angiography on an ARTIS icono system (Siemens) in a pediatric setting to reduce radiation exposure. Contrast administration is limited to less than 2 ml/kg. Typically, this necessitates a 50:50 contrast/heparinized saline dilution for the adequate volume needed for six-vessel acquisition. Right common femoral artery access is obtained under ultrasound guidance with a radial access micropuncture kit to introduce a 5-French radial slender sheath with an outside diameter of 0.021 in. The sheath is positioned halfway in the common iliac artery. We employ live fluoroscopy to prevent iatrogenic injury during sheath advancement. For vessel selection and imaging, we use a 65-cm 5-French Kumpe access catheter (Cook Medical), given the small patient stature.

The right internal carotid artery and superselective angiography of the right anterior cerebral artery revealed a saccular bifurcation aneurysm. Under fluoroscopic roadmap control, a microcatheter was manipulated into the right pericallosal artery aneurysm. Superselective aneurysmography was performed, and embolization was initiated. Unfortunately, given the anatomical location of the aneurysm, it was not feasible to occlude it without disrupting flow to the parent artery. Because it was a distal anterior cerebral artery (ACA) aneurysm, with a known infarct in that distribution, for complete occlusion of the aneurysm and to decrease the chance of recurrence, the decision was made to sacrifice the parent artery by coil and Onyx embolization. Onyx embolic material was administered under the fluoroscopic roadmap. Control angiography demonstrated coil and Onyx embolization of the right pericallosal aneurysm with sacrifice of the parent artery (Fig. 4C–E). Upon placement of the Onyx embolization material, we ensured that our reflux zone was well beyond the area of takeoff of other ACA branches.

The patient tolerated the procedure well, and the postembolization course was uncomplicated. The patient remained without seizures and was able to be extubated. Follow-up MRI revealed a slight increase in the left subdural collection, a stable right subdural collection, and right ACA vascular territory ischemic infarction without evidence of hemorrhagic transformation (Fig. 5). The patient required no further neurosurgical interventions and is currently scheduled for follow-up.

**Patient Informed Consent**

The necessary patient informed consent could not be obtained in this study, because the patient was removed from the custody of the parents due to concerns of nonaccidental trauma. The case was investigated by the proper authorities.
Discussion

Observations

TCAs represent less than 1% of all cerebral aneurysms and result from both penetrating and blunt trauma. They occur more commonly in young patients; Buckingham et al. reported that 30% of aneurysms occur in patients under the age of 20. Interestingly, a male predominance has been reported in the literature, attributed to behavioral predisposition in this age group.

Aneurysm formation is a result of direct vessel injury or stretching of the vessel due to fixation of the vessel to adjacent structures. Distal aneurysms occur predominantly in the ACA as a result of the proximity of vessels to the fixed falx cerebri. Motion of the brain adjacent to the falx can lead to shear forces along the distal ACAs that predispose to traumatic aneurysm formation. This process has been described by several authors. After their formation, TCAs can enlarge rapidly and are at a high risk for rupture. Although the timeline from aneurysm formation to rupture is largely unknown, Fleischer et al. described a 50% rupture rate within 3 weeks of the traumatic event. Importantly, traumatic aneurysms are often pseudoaneurysms due to an absence of collagen layer in the aneurysm wall.

Cases of infantile traumatic distal ACA aneurysms are rare in the literature. Lam et al. described a similar case of infantile trauma in which the pathological examination revealed a traumatic pericallosal artery aneurysm. This patient was managed with direct excision of the aneurysm, sacrificing the parent vessel (ACA). Motohashi et al. reported an infantile pericallosal aneurysm following trauma that spontaneously thrombosed but recurred 6 months later, requiring craniotomy for resection of the aneurysm and involving parent vessel sacrifice. Of note, the authors described the ACA distal to the aneurysm to be vestigial. Levine et al. managed a case of a traumatic infantile pericallosal aneurysm by means of endovascular coiling. In this case, coils were placed within the ACA, embolizing the aneurysm and parent artery. Importantly, in reported traumatic infantile cases of pericallosal aneurysms, parent vessel occlusion is commonly employed. Initial treatment with clipping and coil embolization of the aneurysm alone is unlikely to be successful due to a failure to obliterate the diseased segment of the parent vessel, which can lead to recurrence. In such cases,
parent artery occlusion is a viable method to treat the entirety of the pathology and prevent aneurysm recurrence.\textsuperscript{20,21} In this case, the aneurysm and parent vessel were occluded using coils, with the addition of Onyx embolic material to ensure occlusion of the parent vessel. In our experience, we have not observed aneurysm recurrence or de novo aneurysm formation following adequate parent artery occlusion using Onyx embolic material. According to our review of the literature, this is the first case of traumatic infantile aneurysm embolization using Onyx embolic agent. Our institutional protocol for TCA follow-up includes a clinic visit at 4–6 weeks with head and neck MRA and computed tomography angiography (CTA). The utility of subsequent diagnostic cerebral angiography is predicated on clinical and imaging findings at this appointment.

The outcome in pediatric patients with TCAs is dependent on a variety of factors. Few case reports have described traumatic ACA aneurysms in infants, although sacrifice of the parent vessel is commonly used, as described above.\textsuperscript{6,17,18} Other authors have described endovascular management of TCAs in noninfantile pediatric patients in a series of 15 with TCAs, 8 of which had endovascular management.\textsuperscript{22} There were no instances of aneurysm recurrence or de novo aneurysm formation. In most circumstances, the aneurysm is not an isolated injury, and the presence of parenchymal injury can have a varying impact over time. This patient had multifocal injury to the parenchyma; therefore, the prognosis was uncertain. The degree of injury often correlated with the extent of injury identified on CT and MRI.\textsuperscript{1} There is a paucity of data available on long-term functional outcomes. Extensive brain trauma can have devastating effects on a child, with effects such as developmental delay, motor dysfunction and spasticity, permanent visual and hearing impairment, seizures, and profound intellectual disability. Overall, the prognosis is grim.\textsuperscript{3,23} More than 50% of children ages 0–4 years old with abusive head trauma will die before they turn 21 years old. In surviving children with substantial abusive head trauma, there is a 55% reduction in health-related quality of life.\textsuperscript{1}

Although there are no current guidelines on cerebrovascular injury following NAT, the presence of TCAs following head trauma in the pediatric and infantile population should be considered, given the clinical scenario. Depending on imaging capabilities, initial screening with CTA or MRA can be considered if there is suspicion for cerebrovascular injury. Moreover, if a TCA is identified, proceeding with diagnostic cerebral angiography to evaluate and treat this identified pathology is recommended.

**Lessons**

TCAs are a rare but serious sequela of pediatric abusive trauma. We present a case of a right pericallosal traumatic aneurysm due to abuse that was successfully treated with coil and Onyx embolization of the aneurysm via parent artery occlusion. Rates of pediatric abuse are increasing; thus, TCAs will remain a challenging entity to identify and treat. Despite new methods for the management of this entity, prognosis remains poor due to the diffuse injury often involved in these patients.

**References**


Disclosures
Dr. Spiotta reported personal fees for consulting from Penumbra, Terumo, and RapidAI and stock ownership in AvaI outside the submitted work.

Author Contributions
Conception and design: Hubbard, Cunningham, Saway, Porto, Kosnik Infinger, Spiotta. Acquisition of data: Saway. Analysis and interpretation of data: Saway, Miller, Kosnik Infinger. Drafting the article: Hubbard, Cunningham, Saway, Trano, Miller, Kosnik Infinger. Critically revising the article: all authors. Reviewed submitted version of manuscript: Hubbard, Cunningham, Saway, Miller, Porto, Kosnik Infinger. Approved the final version of the manuscript on behalf of all authors: Hubbard. Administrative/technical/material support: Porto, Spiotta. Study supervision: Hubbard, Porto.

Supplemental Information
Previous Presentations
An abstract of this work was presented as an electronic poster at the 18th Annual Society of Neurointerventional Surgery Meeting in Colorado Springs, CO, July 26–29, 2021.

Correspondence
Zachary S. Hubbard: Medical University of South Carolina, Charleston, SC. hubbardz@musc.edu.