Child's Nervous System (2019) 35:373–378
https://doi.org/10.1007/s00381-018-3968-6

CASE REPORT

Onyx embolization of a ruptured anterior inferior cerebellar artery in a neonate

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Received: 9 March 2018 / Accepted: 31 August 2018 / Published online: 10 September 2018

Abstract

Aneurysmal subarachnoid hemorrhage (SAH) is rare in neonates. The authors present a unique report of a neonate with SAH from anterior inferior cerebellar artery (AICA) aneurysm rupture that was successfully treated with Onyx embolization. This case report demonstrates the utility of Onyx embolization for posterior circulation aneurysms in neonates and the successful management of SAH in this population.

Keywords Subarachnoid hemorrhage · Anterior inferior cerebellar artery · Onyx embolization · Liquid embolic agent · Aneurysm · Posterior circulation

Subarachnoid hemorrhage (SAH) in neonates is rare. While endovascular and microsurgery are the most common treatments for ruptured aneurysms, liquid embolization presents another option for previously inaccessible or difficult-to-treat lesions. We present a unique report of a neonate who suffered SAH secondary to rupture of an anterior inferior cerebellar artery (AICA) aneurysm that was successfully treated with Onyx embolization.

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Case report

History and physical

An otherwise healthy four-day old Hispanic male with an unremarkable birth history was noted to have nausea and diarrhea. The following morning, his mother noticed decreased oral intake and lethargy. Later that day, he developed disconjugate gaze and was admitted to St. Louis Children's Hospital. Upon arrival, the patient was lethargic with sluggish pupils and medial right eye deviation. An initial head CT without contrast (Fig. 1a) demonstrated acute SAH with mass effect on the pons as well as diffuse intraventricular hemorrhage (IVH) and hydrocephalus. CT angiography (CTA) (Fig. 1b) revealed a small right-sided posterior circulation aneurysm. He underwent conventional cerebral angiography after being intubated due to apneic episodes.

Operation

The right femoral artery was accessed using a 4-French sheath. The right vertebral artery was selected with a 4-French JB2 catheter. Initial angiography (Fig. 2a) demonstrated a 1.6-mm fusiform aneurysm arising from a right AICA inferior branch proximally. The diameter of the parent AICA

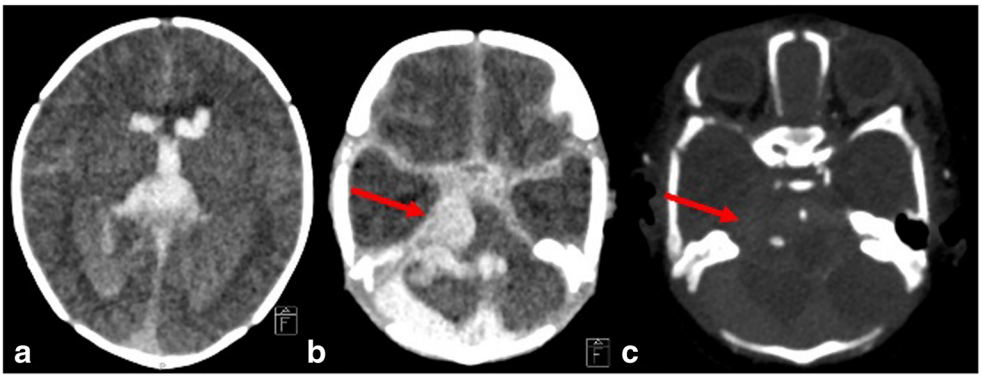

Fig. 1 CT neuroimaging on presentation. **a** Head CT without contrast demonstrating a large amount of subarachnoid hemorrhage within the basal cisterns with diffuse intraventricular hemorrhage and ventriculomegaly. **b** CT demonstrating 4 × 6 × 7-mm hematoma in the right perimesencephalic cistern with mass effect on the pons (arrow). There is blood within the basal cisterns, fourth ventricle, and cisterna magna. **c** CT angiogram windowed for arterial conspicuity demonstrating a small aneurysm in the right perimesencephalic region

at its origin was 0.8 mm before bifurcating into its superior and inferior branches just proximal to basilar artery. A multidisciplinary discussion resulted in a recommendation to pursue endovascular treatment.

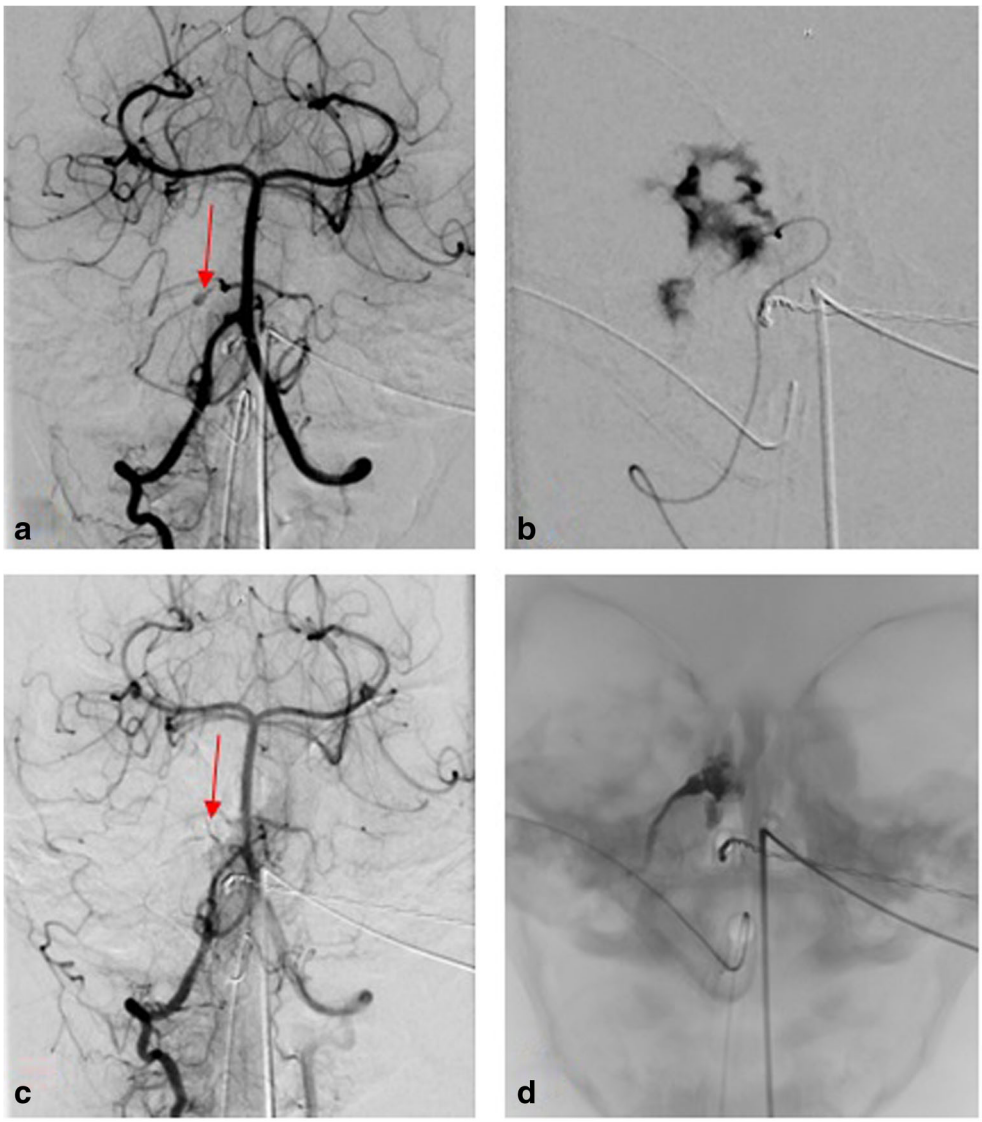

Fig. 2 Cerebral angiography. **a** Right vertebral artery angiogram with magnified frontal view of the posterior fossa demonstrating a 1.6-mm fusiform aneurysm of the right AICA in an inferior branch proximally (arrow), plus decreased cerebellar staining in the right medial cerebellum. **b** Right AICA angiogram with magnified frontal view of the posterior fossa demonstrating contrast extravasation from the AICA aneurysm. **c** Right vertebral artery angiogram with magnified frontal view of the posterior fossa after endovascular sacrifice of the inferior branch of the right AICA by Onyx-34 (Micro Therapeutics, Irvine CA) embolization showing complete opacification of the aneurysm and no evidence of residual extravasation. **d** Frontal view of Onyx-34 (Micro Therapeutics, Irvine CA) cast

A Headway Duo micro-catheter (MicroVention, Aliso Viejo CA) and Transend platinum EX micro-wire (Stryker, Kalamazoo MI) were initially used but we were unable to access the AICA. A second attempt was made with a Synchro-14 soft pre-shaped micro-wire (Stryker, Kalamazoo MI). With a JB2 catheter in the proximal right vertebral artery, the micro-catheter and micro-wire were used to select the inferior branch of the right anterior inferior cerebellar artery.

Immediate selective right AICA angiography through the micro-catheter demonstrated contrast extravasation (Fig. 2b). Embolization with Onyx-34 (Medtronic, Irvine CA) resulted in occlusion of the inferior right AICA branch as well as the small fusiform aneurysm arising from that branch (Fig. 2c). There was Onyx extravasation into the subarachnoid space surrounding the inferior right AICA branch and aneurysm. No Onyx refluxed into the basilar artery. At the completion of embolization, the micro-catheter was withdrawn from the AICA without complication.

Angiography (Fig. 2d) immediately after treatment demonstrated complete obliteration of the inferior branch of the AICA and aneurysm. Anterograde flow in the superior branch of the AICA was maintained. There was no evidence of thrombus formation or distal vessel occlusion. The JB2 catheter was removed, and no further angiography was done due to weight-based contrast dose limitations. Hemostasis was achieved at the femoral artery puncture site after sheath removal using manual compression. The patient was transferred back to the pediatric ICU in stable neurological condition. In total, the infant was exposed to 762 mGy of radiation along with 18 mL of Optiray-320 (Mallinckrodt Pharmaceuticals, St. Louis, MO) contrast administered.

Post-operative course

Immediately after surgery, a head CT (Fig. 3a) revealed diffuse intraventricular hemorrhage with worsening ventriculomegaly; thus, a right-sided extraventricular drain (EVD) was placed. On post-operative day 3, video EEG showed clinical seizures, characterized by left arm posturing and rhythmic arm movement, and subclinical seizures arising from the left frontal and right central head regions. Each seizure lasted 30 s to 5 min and resolved with a single-dose Ativan and increased Keppra dose. The following day (post-operative day 4), the patient experienced five brief (< 30 s) subclinical seizures from the right central regions, which resolved by increasing Keppra to maximum dose. No further seizures were observed in the subsequent 48 h and video EEG was discontinued. Genetic workup during hospitalization was unremarkable.

Surveillance MR angiography (MRA) was performed post-operative day 7 which demonstrated stable aneurysm obliteration and vasospasm in the basilar, and bilateral anterior and middle cerebellar arteries. Permissive hypertension (SBP > 95, or 110% above previous baseline), aggressive hydration, and nimodipine were initiated. Repeat MRA and CT were performed on post-operative day 14 and 18, respectively, and both showed improved clearance of the hemorrhage and attenuation of the vasospasm. Nimodipine was continued for a total of 21 days. The EVD was removed on post-operative day 20 when the CSF had cleared. On post-operative day 23, he developed a fixed medial right eye deviation with inability to close the eye. Head CT (Fig. 3b) showed interval appearance of new intraventricular hemorrhage with blood around the site of the EVD catheter, as well as increased size of the lateral and third ventricles. A 4-cm McComb reservoir was placed the following day and returned xanthochromic CSF. The reservoir was tapped a total of 4 times over 14 days. He was ultimately discharged 3 weeks later after one other episode of seizure that resolved with a single dose of Ativan for a total hospital stay of 46 days. At discharge, our patient had still displayed signs of lateral pontine syndrome. He displayed a right-sided seventh nerve palsy, with a fixed right-eye dilation at 2 mm and his left pupil sluggish. His eyes deviated inward and displayed intermittent horizontal nystagmus. Per audiology, he

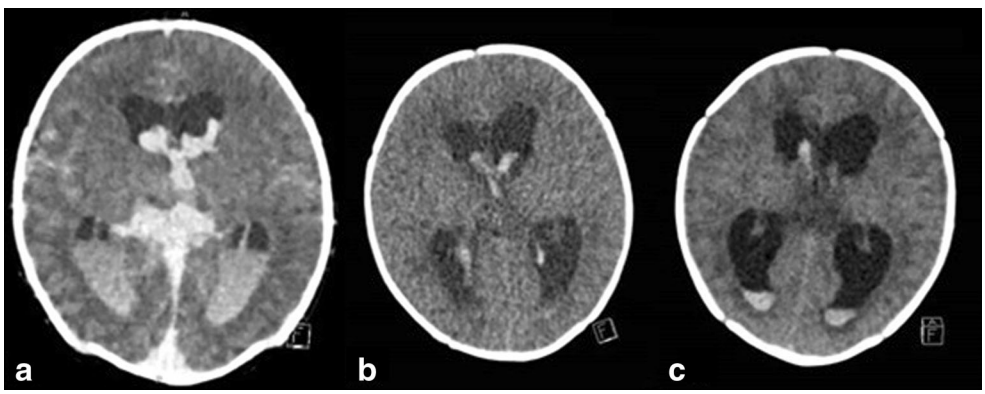

Fig. 3 Post-operative CT neuroimaging. **a** Diffuse intraventricular hemorrhage with subtle slightly worsened ventriculomegaly status post Onyx-34 (Micro Therapeutics, Irvine CA) embolization. There is midline shift to the right thalamus and brainstem due to hematoma. **b** Post-operative day 23, interval removal of right frontal approach EVD with increase in size of both lateral ventricles despite improvement of intraventricular hemorrhage. **c** Post-operative day 27, stable ventriculomegaly following placement of a new ventricular reservoir

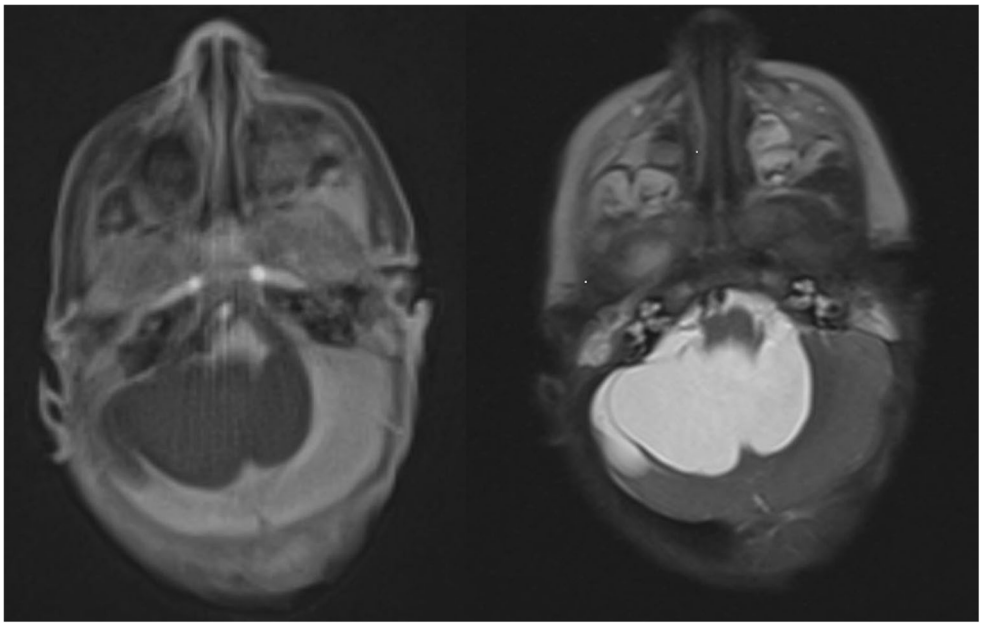

Fig. 4 Follow-up MRI neuroimaging. Fifteen-month follow-up visit with T1- and T2-weighted images (left and right respectively) show interval increase in ventricular dimensions at the level of the medullary-pontine junction. The ventricular catheter was intact with tip in the left lateral ventricle (not shown)

presented with inadequate auditory brain response (ABR) test under natural sleep.

Post-hospital course

Fifteen months after his initial intervention, our patient continues to improve developmentally through stretching with physical therapy. Although he has slight increase in the third and fourth ventricular size with sulcal effacement (Fig. 4), he has been weaned off his ventricular taps. His mother has reported no acute events, or neurologic deterioration. He can now lift his head with his own weight despite still needing minor assistance for head and trunk control. There is noticeable improvement to his lateral pontine syndrome symptoms with increased movements on the right side of his face. However, it still remains flaccid in comparison with his left. Concerned for exposure keratopathy, he underwent a tarsorrhaphy which left him with an eye patch on his eyelid. On neurologic exam, he has symmetric and normal tones in all extremities with his anterior fontanelle noted to be full and bulging. His head circumference has increased to the 80th percentile (pervious 55th percentile). Given these findings, the next course of action is placement of a right occipital ventriculoperitoneal shunt to relieve his hydrocephalus.

Discussion

This is the first reported case of SAH caused by posterior circulation aneurysm rupture repaired with Onyx embolization as well as the first case report of a posterior circulation aneurysm in an infant treated with this technique. Pediatric aneurysms account for less than 5% of all aneurysms and are more likely to occur in males, have fusiform character, and tend to be giant in presentation (> 2.5 cm) [1–4]. Posterior circulation aneurysms account for 17–21% of pediatric aneurysm [1, 3], and within this group 24% are less than 12 months of age [5]. A recent a review of ruptured aneurysms in infants found only two reports of posterior circulation aneurysms [6].

AICA aneurysms account for 1% of all pediatric aneurysms [4]. Mohammad and colleagues reported a partially thrombosed giant basilar artery at the level of the AICA [7] in a 15-year-old boy with sensorineural hearing loss, while another group reported a traumatic intracranial aneurysms in an 8-year-old boy who suffered concurrent arteriovenous malformation after a traffic accident [8]. Thus, to our knowledge, this report serves as a novel presentation of a primary AICA aneurysm rupture in the neonatal population, and the only report of a posterior circulation aneurysm in this population treated with Onyx embolization.

Interventions for pediatric aneurysms have mirrored the adult population with the shifting of the current treatment paradigm to favor endovascular treatments [9]. Endovascular treatment is less invasive and allows for access to difficult-to-expose aneurysms [10]. While some institutions have reported a higher complete obliteration rate with microsurgery versus endovascular treatment [11], analysis of the Kids' Inpatient Database (KID) demonstrated a higher hospital mortality, complication rates, and overall costs of surgical clipping when compared to endovascular modality [12].

Within the realm of endovascular interventions, traditional methods (coil, stenting, flow diversion) have required larger and less flexible catheters that are inaccessible to smaller and distal aneurysms, which may result in rupture of nearby vasculature [13]. Liquid embolization offers a unique solution to

this problem, as well as the potential to fill the aneurysm completely [14]. *N*-butyl cyanoacrylate (nBCA) is preferred by some operators but it can have issues with the delivery precision due to its adhesiveness to ionic compounds, such as blood [15]. Furthermore, potential mistiming of embolization with an nBCA could result in reflux into the basilar artery lumen, which would highly complicate the case. The Onyx liquid embolic system was developed as a nonadhesive liquid composed of ethylene vinyl alcohol copolymer (EVOH) dissolved in dimethyl sulfoxide (DMSO) with tantalum precipitate. As the Onyx solution is injected, the DMSO will wash away in the vasculature while the EVOH and tantalum form a polymeric embolus [16]. Significant disadvantages of using this intervention are operator dependence, learning curve, costs, and inability to be redacted [15]. The pediatric population presents a particular issue with a higher susceptibility to toxicity due to relatively low weight. Within the literature, Onyx has been used with success in the pediatric population with infectious intracranial aneurysms, traumatic intracranial aneurysms, pseudoaneurysms, and arteriovenous malformations (AVMs) [14, 17–19]. In the adult population, Onyx has been used to treat a variety of pathologies, including an AICA flow-dependent aneurysm [20].

The AICA is small in diameter and highly variable in its course, which made the technical aspect of endovascular treatment rather difficult. The AICA may branch from the vertebral, basilar, or posterior inferior cerebellar artery (PICA) [21]. An AICA that is strongly developed has been defined as > 1.2 mm in an adult brain [22]; thus, in a pre-mature neonate, the vessel would be expected to be far less developed and more sensitive to endovascular shear forces. The risk of hemorrhage related to catheterization of very small vessels of the cerebral vasculature is ever present. The extravasation of contrast seen in our patient could have been from the dissection of a fragile AICA or re-rupture of the AICA aneurysm and likely not from wire perforation, as the wire was seen to follow the lumen of the vessel on roadmapping. The decision to use a micro-catheter with a smaller distal tip OD may have been considered but would not necessarily have altered outcomes. Selecting the 0.53-mm distal tip OD Headway Duo (MicroVention, Aliso Viejo CA) as the micro-catheter of choice preserved the option of coil embolization in addition to liquid embolic agent embolization. The use of other catheters, which may have a smaller distal tip OD, would have been a viable technique for an nBCA embolization only. However, the operators decided that having the flexibility of treatment options using the Headway Duo (MicroVention, Aliso Viejo CA) microcatheter provided outweighed the value in treating this particular patient than the slight decrease in distal tip OD achieved by other less flexible catheters and nBCA.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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