pediatric stroke

Ruptured Intracranial Arterial Aneurysm in a Neonate

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Abstract

Hemorrhagic stroke accounts for half of incident strokes in children yet remains understudied. Clinical and research teams assessing recovery from pediatric hemorrhagic stroke are limited because commonly used outcome measures may not be meaningful to the care team, educators, and foremost the child and family. The current paper serves as a model for bringing together multiple disciplines for shared purposes of better assessing child recovery and development after hemorrhagic stroke over time, advocating for child- and family-centered care, and facilitating discourse in the community of those caring for children with pediatric hemorrhagic stroke. The purpose of this work is to describe our guiding principles and the process of our team's development of a toolkit that emphasizes inclusion of the patient and family perspective of recovery in the home, school, and community contexts. We provide the toolkit and an example of specific outcome measures selected to address unique aims of a research project. The toolkit is intended to be a living platform for further evolution of outcome measures for pediatric hemorrhagic stroke. As such, we hope it will be a dynamic document that serves the ongoing and future clinical and research needs of the multidisciplinary professional teams providing holistic care after a pediatric hemorrhagic stroke.

Introduction

Intracranial aneurysms (IA) are quite common in the world population, with a prevalence of approximately 4% (1). They are defined as focal abnormal dilatations of intracranial blood vessels. In most of the cases these abnormalities are small and asymptomatic, being diagnosed as incidental findings or during screening tests in patients with a predisposing genetic disorder or relatives with a known IA. Only 10-15% of them are symptomatic, mainly owing to compression when there is a large CA, microbleeds that irritate the meninges or when there is a rupture with subdural hemorrhage (1).

Pediatric cerebral aneurysms are rare compared to adults. It is less than 4%.(3) The first case report of aneurysm in pediatric population was described by Eppinger, a German pathologist, in the nineteenth century, on autopsy of a 15 years old child who died of aortic stenosis. (4)The incidence in infants younger 1 year is extremely low; it is height in adulthood (0.52/100000 person/year) compared with children younger 4 years old (0.006/100000 person/year). (5) Goia mentioned in their paper about 18 cases of neonates with aneurysm until 2020. In the pediatric population the factors that contribute with aneurysm are: trauma, infection, moyamoya disease, fibromuscular dysplasia, sickle cell anemia, coarctation of the aorta, arteriovenous malformation, polycystic renal disease, collagen disease; and if

known that the most common pediatric cerebral aneurysm are caused after head trauma. (6)

We report a patient with cerebral aneurysm in the neonatal period and that it was thrombosed itself.

Case Description

A 14-day-old premature boy was admitted with hypotonia, lethargy, and vomiting. The patient was born at 35 weeks´gestation through cesarean section, in the setting of triplet pregnancy.

The patient was suspected of having sepsis so blood, urine and cerebrospinal fluid (CEF) tests were run. The CEF analysis showed red blood cells, therefore a transfontanellar ultrasound was performed, revealing intraventricular hemorrhage. (Fig 1)

Figure 1. Transfontanellar ultrasound. A) First ultrasound performed 6 days after the symptoms began. The coronal plane shows the atrium of the lateral ventricles, which are enlarged (14 mm each), and intraventricular hemorrhage grade III. B) Ultrasound 15 days after the initial evaluation. The coronal plane shows a hypoechoic round structure in the midline, suggesting the third ventricle. C) With color Doppler, the hypoechoic structure displays prominent flow, indicating proximity to the anterior cerebral artery.

On the 9th day the MRI (Fig. 2) revealed ischemic lesions in both ACA territories, interhemispheric and suprasellar subarachnoid hemorrhage, with intraventricular extension and cortical superficial siderosis. However, the source of these findings was unknown yet. Furthermore, the arterial structures in the TOF sequence were difficult to evaluate due to the presence of blood remains, which is why a computed tomography angiography (CTA) of the brain was requested 6 days later (day 15 after the onset of symptoms).

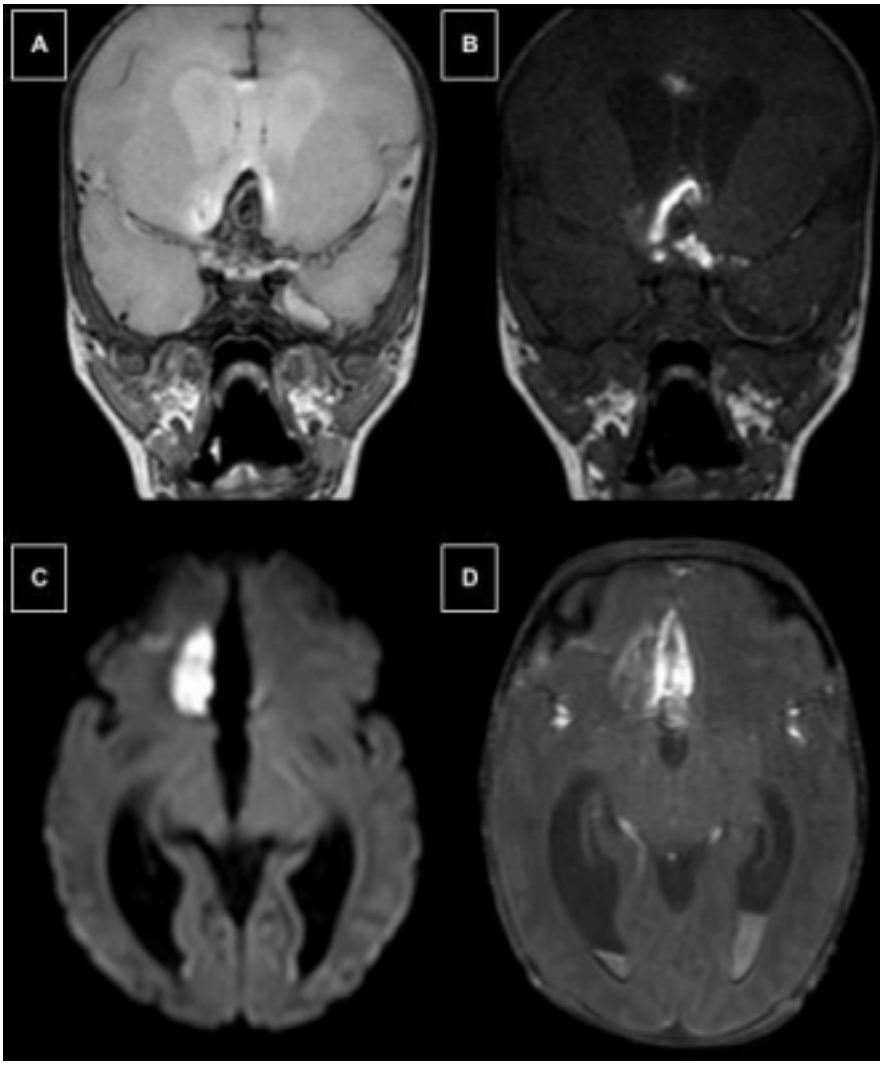

Figure 2. MRI of the brain. A) Coronal T2: subarachnoid hemorrhage in the antero-inferior interhemispheric region, with a slight mass effect on the medial aspect of both frontal lobes. B) Coronal T1: hyperintense signal of the subarachnoid hemorrhage, consistent with the subacute stage. C) DWI: restricted lesion in the right medial frontal lobe, compatible with acute ischemic stroke in the anterior cerebral artery territory. D) TOF: interhemispheric subarachnoid hemorrhage obscures the proper assessment of the anterior cerebral arteries.

The CTA showed a saccular aneurysm located in the left anterior cerebral artery at the origin of the anterior communicating artery (Fig 3). No procedure was performed after this. Three weeks later another CT Angiography revealed spontaneous thrombosis (Fig 4). No treatment was needed.

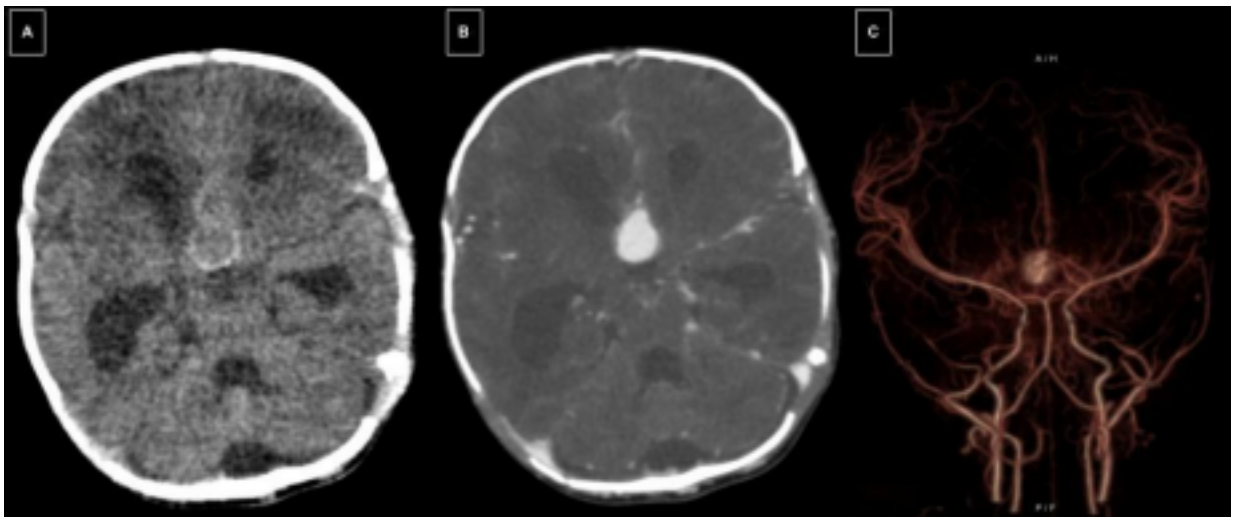

Figure 3. CTA. A) Noncontrast axial image. It is observed an hypoattenuating nodule in anterior arterial topography, between A1 and A2 segments, a finding that suggests an aneurysm. B) The same plane with contrast shows that the nodule enhances and presents an irregular rounded shape. C) 3D reconstruction CT. Left anterior cerebral artery saccular aneurysm is confirmed.

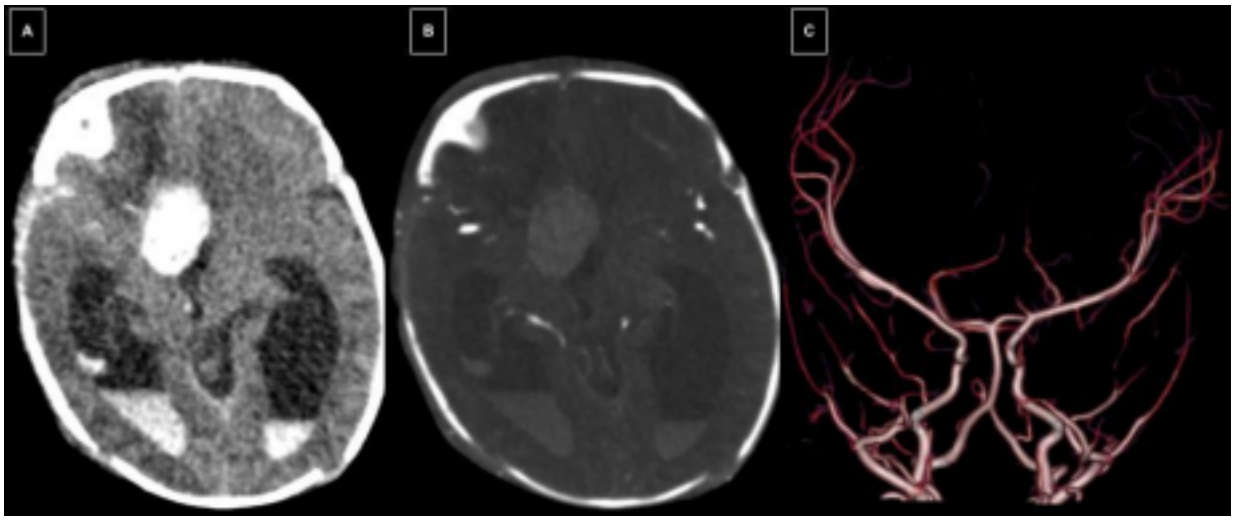

Figure 4. CTA. A) Noncontrast axial image. Enlarged hyperattenuating interhemispheric structure associated with similar findings in both ventricles, which was interpreted as remaining blood products. B) Axial image with contrast. Lack of enhancement of the aneurysm after the administration of intravenous iodinated contrast, finding that means it is thrombosed. C) 3D reconstruction CT. There are no signs of the aneurysm seen in the prior CT (Fig 3)

Radiological Findings

Transfontanellar cranial ultrasonography performed on the sixth day of admission showed a bilateral intraventricular hemorrhage with dilated ventricles.
On the ninth day of admission contrast enhanced MRI confirmed extensive intraventricular and subarachnoid hemorrhage, but the cause was unknown.
Brain CTA revealed left ACA saccular aneurysm and transfontanellar Doppler ultrasonography performed on the fifteenth day of admission showed an intracranial rounded hypoechogenic structure near the anterior cerebral artery with a double bidirectional flow. Three weeks after the first CTA was performed, a second one showed spontaneous thrombosis and complete exclusion of the aneurysm.

Discussion

Pediatric brain aneurysms are a rare pathology but they are exceptional in neonatal patients with a male predominance (7). The pediatric population accounts for less than 5% of all cerebral aneurysms. (6,8,9) It is known that the risk factors are different from adults to children due a children do not have risk factors some: age, hypertension, smoking, diabetes, hypercholesterolaemia and alcohol. However, in children there are other risk factors: cerebral arteriovenous malformations, connective tissue disorders, sickle cell anemia, polycystic kidney disease. (4) (8)(6) There are 8 different pathogenic subcategories, being idiopathic aneurysms the most common ones, followed by traumatic aneurysms and those due to excessive hemodynamic stress. (10)

Clinical presentation is extremely variable. The more frequent presentation is with hemorrhage: subarachnoid hemorrhage. (8)(11) Common clinical manifestations are seizures, alteration of consciousness, irritability and bulging fontanelle (12,13).

Non-invasive transfontanellar ultrasound is an attractive first-line imaging option for identification of large hemorrhage. Most patients receive MRI and CT Angiography to diagnose intracranial hemorrhage and the cause. Cerebral angiography remains a gold standard technique, but due to its high risk it is not the first investigation in neonates. (12)The form of aneurysm in children is bigger than in adults.

Only 5-10% present spontaneous thrombosis (10).

In a systematic review published in 2020, reviewing the bibliography from 1980 to 2020, considering only aneurysm without association with another vascular malformation, they included 88 articles of which 75 were case reports and 13 articles were from small series (between 2 to 15 patients).(8) All of patients were less than 2 years all (infants). (8) The occurrence of IA in neonates is consequently exceptionally rare with only 17 cases reported in the last 20 years. (5)(14)

In our case, the child has a mild developmental delay. Slator published in a British serie that 44% of

children who survived had neurological sequelae at 1 year, which included seizures, cerebral palsy and behavioral difficulties. (4)

The mortality rate was 12.4% and the only predictor of poor neurological outcome was mass effect. (8)

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