Ruptured Intracranial Aneurysm in a Neonate: Case Report and Review of the Literature

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Key words
- Clipping
- Embolization
- Intracranial aneurysm
- Neonate
- Subarachnoid hemorrhage

Abbreviations and Acronyms
CT: Computed tomography
IA: Intracranial aneurysm
MCA: Middle cerebral artery

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BACKGROUND: **Intracranial aneurysms (IAs) are exceptional in neonates accounting for less than 2% of all IAs occurring during the first decade of life. Little is known about this pathology in this specific population. Because of its scarcity and this specific age at onset, the treatment of IA in neonates is challenging. We describe a rare case of aneurysmal subarachnoid hemorrhage in a neonate and review the current literature.**

CASE DESCRIPTION: **A 21-day-old boy was admitted for hypotonia, vomiting, and seizures. Computed tomography scan revealed a subarachnoid hemorrhage in the sylvian fissure, a frontoparietal subdural hematoma, a left middle cerebral artery (MCA) aneurysm with a diameter of 11 mm, and an infarct of the MCA frontal region. He was successfully treated with endovascular coiling, neuroprotection, and antiepileptic drugs. Immediate postoperative magnetic resonance imaging showed a good aneurysm occlusion without any further ischemia. The outcome was favorable with extubation at day 10. At follow-up, the child experienced normal psychomotor development with no motor deficit.**

CONCLUSIONS: **Ruptured IAs in neonates are rare. Subarachnoid hemorrhage is the most common presentation. Intracranial aneurysms are frequently larger than 10 mm and located on the MCA. The treatment could be surgical or endovascular depending on the characteristics of the aneurysm. There is no recommendation concerning the prevention or treatment of vasospasm in neonates.**

INTRODUCTION

Intracranial aneurysms (IAs) are uncommon in the pediatric population, accounting from 0.5% to 4.6% of all IAs.[1,2] The incidence of IA is highest in late adolescence (0.52 per 100,000 person-years) compared with the 0- to 4-year old age group (0.06 per 100,000 person-years).[3] The occurrence of IA in neonates is consequently exceptionally rare, with only 17 cases reported in the last 20 years.[4-20] Because of its scarcity, we have very few data concerning IA in this population. Moreover, risk factors of IA are completely different from the adult population and its pathogenesis cannot be extrapolated from adult series. In the pediatric population, the onset of IA is more frequent in boys and is associated with collagen mutation diseases such as polycystic kidney disease, fibromuscular dysplasia, Ehlers-Danlos syndrome, or Marfan syndrome.[6,21,22] The type of IA is also different with a higher prevalence of nonsaccular, giant IAs.[23,24] Therefore, the treatment of IA in children and neonates is challenging with technical issues leading to a high risk of morbidity, and we have no clear recommendations for the management of these patients. Herein, we report an exceptionally rare case of a ruptured IA in a 21-day-old neonate and present a current review of the literature.

CASE REPORT

A 21-day-old white boy was admitted in our institution for sudden-onset hypotonia and vomiting. He was born at term and had no history of trauma or infection. The initial examination showed hypotonia, right hemiparesis, and seizure. A left anisocoria was noted. He rapidly presented in a comatose state. It was decided to intubate him and an antiepileptic medication was initiated. A computed tomography (CT) scan showed a left Fisher grade IV subarachnoid hemorrhage mainly located in the sylvian fissure with a left frontal subdural hematoma and frontal ischemia. A contrast-enhanced CT scan (Figure 1) was performed to define the etiology of the bleeding and revealed an 11-mm aneurysm on the left middle cerebral artery (MCA) superior (frontal) branch right after the MCA trifurcation and a vasospasm of the left MCA. Because of his young age and after a multidisciplinary discussion, it was decided to treat him by endovascular means. The endovascular procedure was successful and consisted in simple coiling (Figure 2), leading to a complete exclusion of the IA. The superselective injections from the microcatheter showed that its development incorporated the origin of the prefrontal branch bifurcation (Figure 3), which further suggests a

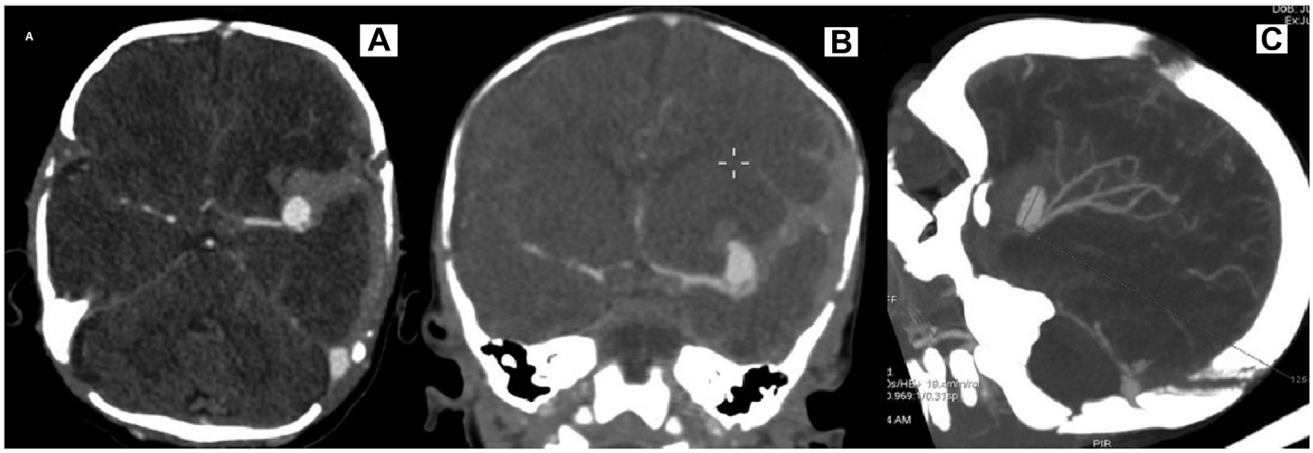

Figure 1. Contrast-enhanced computed tomography scan showing a left middle cerebral artery ruptured intracranial aneurysm in axial (**A**), coronal (**B**), and sagittal view (**C**).

dissecting origin. Regarding the treatment of vasospasm, it was decided to monitor the cerebral perfusion pressure and intracranial pressure with daily transcranial Doppler ultrasound. Because of the absence of data in pediatric patients, we did not use nimodipine. At day 2, postoperative magnetic resonance imaging scan (Figure 4) showed an ischemic stroke in the superficial frontal left MCA territory, and a decision to prolong leading to neuroprotection was taken. The outcome was favorable with extubation at day 10. He was discharged home at day 21 with no motor deficit. At 8-month follow-up, psychomotor development was normal.

LITERATURE REVIEW

The PubMed/MEDLINE database was used to perform an online search of reported studies using the following key words and Medical Subject Heading terms: "neonate," "intracranial," "aneurysm," "cerebral," and "paediatric." Only full-text articles in English were chosen. Articles were selected from January 2000 to January 2020 to offer a recent up-to-date review of the literature.

According to the best match and relevance, the search results were refined. Only cases of spontaneous IA in neonates

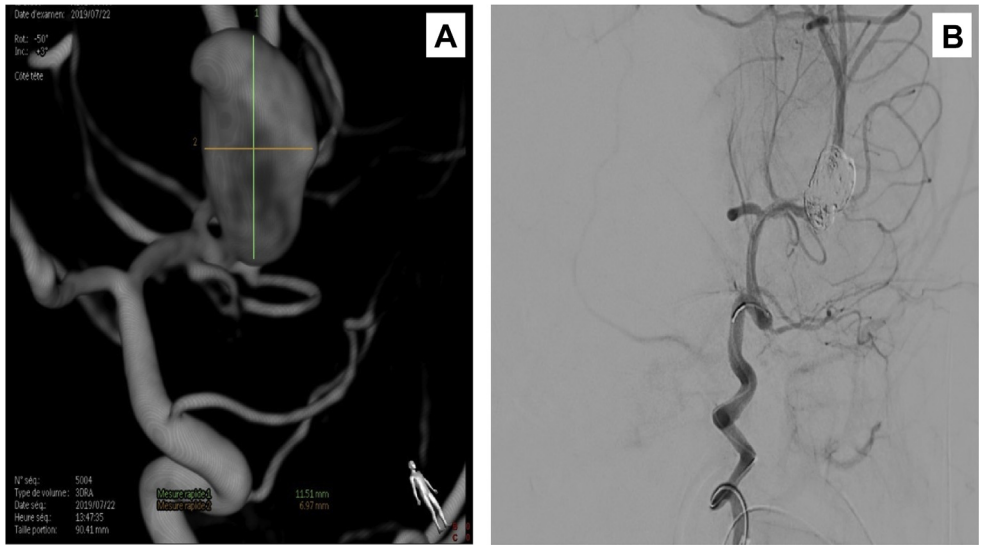

Figure 2. Digital subtraction angiography showing the 11-mm aneurysm located on the left middle cerebral artery on 3-dimensional reconstruction (**A**) and 2-dimensional images after coiling and complete occlusion (**B**).

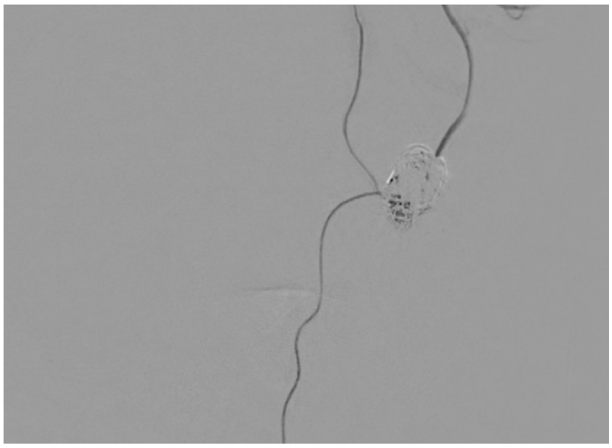

Figure 3. Digital subtraction angiography with superselective injections from the microcatheter showing intracranial aneurysm development incorporating the origin of the prefrontal branch bifurcation.

were included. Mycotic IA, traumatic IA, or aneurysm associated with arteriovenous malformation were excluded from the review. After applying the aforementioned criteria, the literature review yielded 18 IAs in 17 articles. The selected cases of neonatal IA are presented in Table 1.

DISCUSSION

In this study, we report a very uncommon neonatal IA presenting with subarachnoid hemorrhage and present a review of the literature of the last 20 years (Table 1). The mean age of onset of IA in neonates is 20.9 ± 8.9 days with a discrete female predominance (9 girls vs. 7 boys). Seizure in symptomatic IA is the main presentation (10 out of 18 patients) followed by intracranial hypertension. In a recent study focusing on the risk of seizure in pediatric patients with IA,[16] it appeared that pediatric patients are at higher risk of seizure in case of subarachnoid hemorrhage compared with the adult population. The advanced hypothesis for this is the vulnerability of the cortex in pediatric patients[25] and the increased prevalence of IA located in the MCA associated with cortical hemorrhage.[16,26] Another issue of managing IA in the neonates is the diagnostic modalities. In a former study, Tekkök and Ventureyra[27] proposed cranial ultrasound as a first step diagnostic modality. We have observed in our recent review that if cranial ultrasound was used to assess the presence of hydrocephalus or monitor the brain suffering, contrast-enhanced magnetic resonance imaging or CT scan was predominantly proposed to establish a clear etiologic criterion of IA.[14,15] In the present case, we think angiography should be proposed only in the intention to perform an endovascular treatment. Angiography is an invasive diagnostic modality and technical constraints exist such as the need for adapted catheters or the quantity of iodine contrast administered during the endovascular procedure.[17] Concerning the IA characteristics, the main location was MCA (11/18) followed by anterior cerebral artery (4/18) and internal carotid artery (2/18) (Table 1). Aneurysms were frequently nonsaccular (7 vs. 6 with the available data) with a mean diameter of 10.5 ± 6.9 mm. We observed in pediatric patients a greater mean diameter compared with adult patients where more than 50% of the patients presented with a mean IA diameter of 5 mm or less.[28] Risk factors of IA in children are different from adult population where high blood pressure, smoking, and age are the most common.[28] Because of its scarcity, data concerning IA in neonates are based on case reports, and risk factors are not clearly represented. Head trauma, infectious IA, dissecting IA, and collagen mutation disease are mentioned in the literature.[23] Concerning therapeutic options, microsurgical clipping[8] or endovascular approach[7] can be applied (available data). The decisive factors for the treatment choice are as follows: the experience of each professional, IA location, and the availability of adapted devices such as surgical clips or endovascular catheters. The lack of data for these patients does

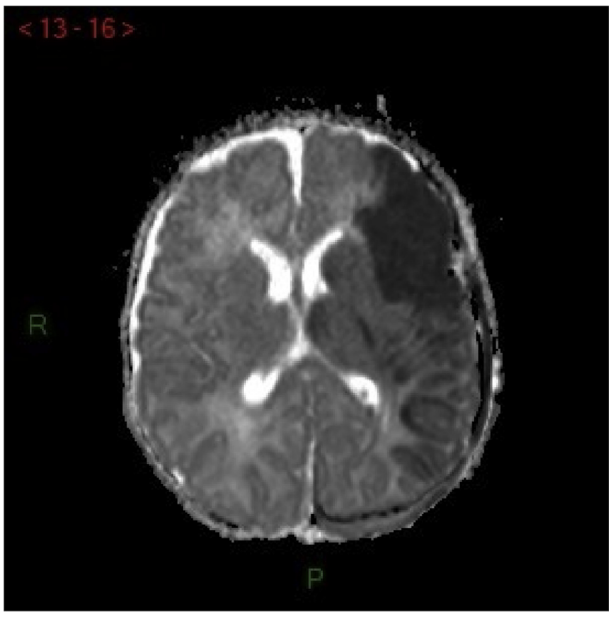

Figure 4. Axial view of diffusion-weighted magnetic resonance imaging scan showing ischemia on the middle cerebral artery territory.

Table 1. Literature Review of all Studies Reporting Neonatal Intracranial Aneurysm from 2000 to 2020

Study	Number of Patients	Sex	Age	Clinical Presentation	IA Location	IA Morphology (Saccular/ Nonsaccular)	IA Size	Treatment Modalities	Outcomes
Maroune et al.[19]	1	M	3 days	Seizures, irritable, cyanotic	MCA	Saccular	10 mm	Microsurgical clipping	Vegetative, death at 4 years follow-up
Motohashi et al., 2013[9]	1	F	28 days	Seizures, vomiting, fever, anterior fontanelle bulging	ACA	X	7 mm	Microsurgical clipping	Complete recovery
Gallia et al., 2005[11]	1	F	28 days	Left eye ptosis	ICA	Nonsaccular	20 mm	Embolization	Complete recovery
Lasjaunias et al., 2005[6]	2	F	8 days, 28 days	Fever, meningitis/ anterior fontanelle bulging	ACA, MCA	1 saccular/1 nonsaccular	X	Microsurgical clipping	Complete recovery
Song et al., 2005[10]	1	F	11 days	X	ACA	Nonsaccular	X	Embolization	Incomplete recovery
Rana et al., 2007[5]	1	M	21 days	Seizures, hemiparesis	MCA	Saccular	5 mm	Embolization	Complete recovery
Wong and Fong, 2007[14]	1	M	28 days	Seizure	MCA	X	5 mm	Surgical excision and anastomosis	Complete recovery
Kasliwal et al., 2008[20]	1	M	21 days	Seizure, irritability	ICA	Nonsaccular	26 mm	Spontaneous ICA thrombosis	Complete recovery
Van Raay et al., 2009[15]	1	F	7 days	Fever, axial hypotonia, irritability, anorexia	PICA	Nonsaccular	5 mm	Microsurgical clipping	Complete recovery
Vizcaino et al., 2009[18]	1	M	26 days	Anterior fontanel bulging, seizure	MCA	Saccular	20 mm	Scheduled embolization at 40 days	Death before embolization
Tai et al., 2010[12]	1	M	9 days	Seizures, fever, anterior fontanelle bulging	PICA	Nonsaccular	3 mm	Embolization	Complete recovery
Choi and Lee, 2013[8]	1	X	26 days	Hemiparesis, bulging anterior fontanel	MCA	Saccular	10 mm	Stereotactic hematoma aspiration	Rebleeding, surgical clipping
Rao et al., 2013[7]	1	F	21 days	Seizure, hemiparesis	MCA	Nonsaccular	8 mm	Embolization	Global development delay
Kim et al., 2014[4]	1	F	28 days	Anemia	MCA	X	7 mm	Microsurgical clipping	X
Yatomi et al., 2014[17]	1	F	28 days	Seizure	ACA	X	X	Embolization	Complete recovery
Mohotti et al., 2018[13]	1	F	28 days	Seizure, anterior fontanelle bulging	MCA	X	14 mm	Microsurgical clipping	Persistent hemiparesis, seizures
Chen et al., 2018[16]	1	M	28 days	Seizure, coma	MCA	X	X	X	Incomplete recovery
Present study	1	M	21 days	Coma, seizure, anterior fontanelle bulging	MCA	Saccular	8 mm	Embolization	Complete recovery

IA, intracranial aneurysm; M, male; MCA, middle cerebral artery; F, female; ACA, anterior cerebral artery; X, not specified; ICA, internal carotid artery; PICA, posteroinferior cerebellar artery.

not allow a decision-making algorithm for the treatment of these patients and a case-by-case decision is based on clinical situation and IA morphology. Concerning the risk of recurrence, a recent study[29] estimated an annual aneurysmal recurrence rate of 2.6%. The only determinant for recurrence was IA greater than 5 mm. Outcomes in the literature review appeared favorable in 10 of 18 patients and 2 patients died during management or at follow-up. We cannot compare the data from 18 patients with adult cohorts whose mortality rate was around 7% in treated patients with 20% of the patients dying before reaching hospital.[28] In a recent review of pediatric intracerebral hemorrhage,[30] it appeared that the mortality rate was lower in the pediatric population compared with adulthood and its main determinants were the hemorrhage volume, the occurrence of hydrocephalus or brain herniation, and the early altered mental status. Long-term outcomes of IA in the pediatric population have been described recently in 2 large studies from the same team.[31,32] With a mean follow-up of 25 years, the authors have noted a long-term excess mortality in pediatric patients

presenting with IA compared with the general population. Prognosis was worse in boys' population, in case of ruptured IA at onset, and when subarachnoid hemorrhage occurred before 15 years of age. In the 1-year survivors after subarachnoid hemorrhage, causes of death were a recurrent SAH in 10% because of de novo IA or recurrence of the treated IA. Long-term outcomes appeared favorable in 62% of the patients, 35% died, and 3% remained dependent. The factors associated with good outcomes were a good clinical status at admission, IA from the anterior circulation, absence of vasospasm, and good IA exclusion.[32] The type of treatment did not seem to affect long-term outcomes in this study.

CRediT AUTHORSHIP CONTRIBUTION STATEMENT

Alice Goia: Data curation, Formal analysis, Writing - review & editing. **Elisabeth Garrido:** Data curation, Formal analysis, Writing - review & editing. **Margaux Lefebvre:** Data curation, Formal analysis, Writing - review & editing. **Olivier Langlois:** Writing - original draft, Writing - review & editing. **Stéphane Derrey:** Writing - original draft, Writing - review & editing. **Chrysanthi Papagiannaki:** Writing - original draft, Writing - review & editing. **Vianney Gilard:** Conceptualization, Methodology, Writing - original draft, Writing - review & editing.

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