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Ruptured Middle Cerebral Artery Aneurysm in an Infant: Case Report and Literature Review

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Abstract

Intracranial aneurysms (IA) in infants are reportedly rare at 0.5% to 4.5% of all aneurysms. Furthermore, subarachnoid hemorrhage in infants younger than three months are even rarer as it has been reported in approximately 20 cases only till date.
A 3-month-old infant with seizures and impaired consciousness was admitted to our hospital. Three-dimensional computed tomography angiography (3D-CTA) revealed a dissecting aneurysm with a maximum diameter of 13 mm in the right M2. Internal trapping using detachable coil were successfully performed, following which he was discharged without significant neurological deficit after one month of onset. Thus, we have reported a rare case of a large ruptured dissecting IA in a 3-month-old infant, in the right middle cerebral artery (MCA), successfully treated with an endovascular therapy, along with a literature review.

Keywords: aneurysm, infant, subarachnoid bleeding, subarachnoid hemorrhage, endovascular treatment

Introduction

Intracranial aneurysms (IAs) in infants are rare, reportedly accounting for 0.5-4.5% of all aneurysms.[1-5] We report a case of subarachnoid hemorrhage due to a ruptured aneurysm in the right middle cerebral artery (MCA) that was morphologically identified as a dissecting aneurysm in the literature. Consent for submission of the paper was obtained from the patient.

Case Report

A previously healthy 3-month-old male infant with an uncomplicated childbirth, no family history of cerebrovascular disease, and no known trauma, was admitted to the peripheral hospital after an episode of seizure and impaired consciousness (Glasgow Coma Scale 12; E4 V2 M4). He underwent head computed tomography (CT), which revealed a subarachnoid hemorrhage (SAH) (Fig. 1). Three-dimensional computed tomography angiography (3D-CTA) revealed an aneurysm with a maximum diameter of 13 mm in the peripheral right MCA the next day, following which he was referred to our hospital for treatment under sedation. Clinical examination detected pulsations in the anterior fontanelle which was slightly tensed, and both the pupils to be 3.5 mm in size, with p. Initial computed tomography (CT) showed an SAH centered on the right sylvian fissure. 3D-CTA indicated a dumbbell-shaped saccular aneurysm with a maximum diameter of 13 mm on the right M2. Cerebral angiography was performed under general anesthesia. The right internal carotid artery (ICA) angiogram showed that the maximum diameter of the aneurysm was 5.4 mm, which was smaller than that on 3 D-CTA. Consequently, based on the shape and 3D-CTA findings, it was considered to be associated with thrombosis in the false lumen or aneurysm. On the basis of the aneurysm branching anteriorly to the right frontal lobe from the tip (Fig. 1), we diagnosed it as a right M2 dissect-

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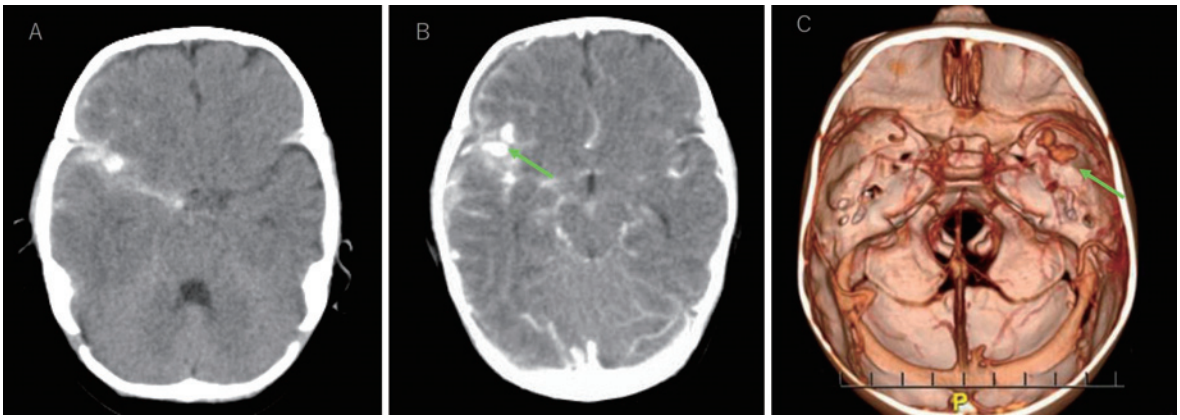

Fig. 1 Axial section of CT (A) showing a SAH centered on the right Sylvian fissure. 3D-CTA (B) and (C) showed a dumbbell-shaped saccular aneurysm (arrow) with a maximum diameter of 13 mm in the right MCA.

ing aneurysm.

Clinical course and endovascular surgery

Using stent-assisted coil embolization or flow-diverter stents is typically considered a method to preserve the patency of the parent artery. However, in Japan, these reconstructive treatments using a stent are off-label during the acute ruptured phase; in addition to the limited diameter of the M2 vessel of less than 1 mm and the absence of widely accepted dual antiplatelet therapy for pediatric patients, a durable deconstructive treatment was deemed desirable for pediatric patients. A previous study reported that internal trapping without bypass was performed successfully in children beyond M2,[6] and the branch from the aneurysm was anterior to the right frontal lobe; therefore, we planned internal trapping using detachable coil under general anesthesia. A 22-gauge intravenous catheter needle was inserted into the right femoral artery, and a 3 Fr short sheath was inserted and replaced with a Flexor Shuttle Guiding Sheath 4 Fr 90 cm (Cook Medical Inc., Bloomington, IN, USA) using a 0.035-inch Radifocus Guide Wire (Terumo, Tokyo, Japan). Additional intravenous heparin was not administered because the patient was heparinized using saline irrigation. The shuttle sheath and Berenstain catheter were coaxially assembled and a guiding catheter was placed in the right ICA, confirming the presence of an aneurysm on the right ICA angiogram. Balloon occlusion test was difficult due to the small diameter of the vessels, and confirmation of leptomeningeal anastomosis was abandoned. We attempted to advance the microcatheter Excelsior SL-10 (Stryker, Kalamazoo, MI, USA) into the aneurysm over a Tenrou 1014 micro guidewire (Kaneka Medix, Osaka, Japan). As the M2 spasm was so severe that the catheter could not be advanced sufficiently, the microcatheter was replaced with a Marathon catheter (Covidien, Irvine, CA, USA) and placed in the aneurysm. Eight coils were implanted with ED coils (Kaneka Medix) to achieve complete occlusion of the parent vessel (Fig. 2). In total, the pediatric patient was subjected to a cumulative radiation exposure of 301 milligray (mGy) and received an administration of 16.75 milliliters (mL) of contrast media.

Postoperative course

Postoperatively, intubation was continued under sedation for several days. A CT scan on the day after surgery showed a low-density area in the right frontal lobe, consistent with an occluded area. The hematoma did not increase in size (Fig. 2). Symptomatic epilepsy, probably due to cerebral vasospasm, was observed the day after surgery, and levetiracetam (30 mg/kg) was initiated. Thereafter, the patient was treated for cerebral vasospasm and discharged home one month after the onset of the disease, with adequate postoperative care and no gross paralysis. One year after the surgery, 3D-CTA and Magnetic Resonance Imaging (MRI) evaluation revealed no significant right M2 findings, and the cerebral tissue around the aneurysm had encephalomalacia (Fig. 3). No radiographic deformity of the coils was visible post-surgery. The Developmental Quotient (DQ) using the Kinder Infant Development Scale was 100, and his development as a one-year-old was very slightly delayed. Genetic testing of the connective tissue performed after hospital discharge however, showed no abnormalities. We plan to continue follow-up with an MRI every year.

Discussion

Pediatric IA is rarer than adult IA, especially in infants. Not much is known about its etiology. Compared to adults, they tend to be more fusiform than saccular, larger, and more irregular in shape.[7] Lasjaunias et al. suggested that they are congenital, arising due to various vessel wall dysfunctions and repair processes.[8] The etiology of pediatric IAs is identified in less than half of the cases; the breakdown of that is as follows: about 50% have vascular dissection, 15% have infections, and 5-10% are associated with

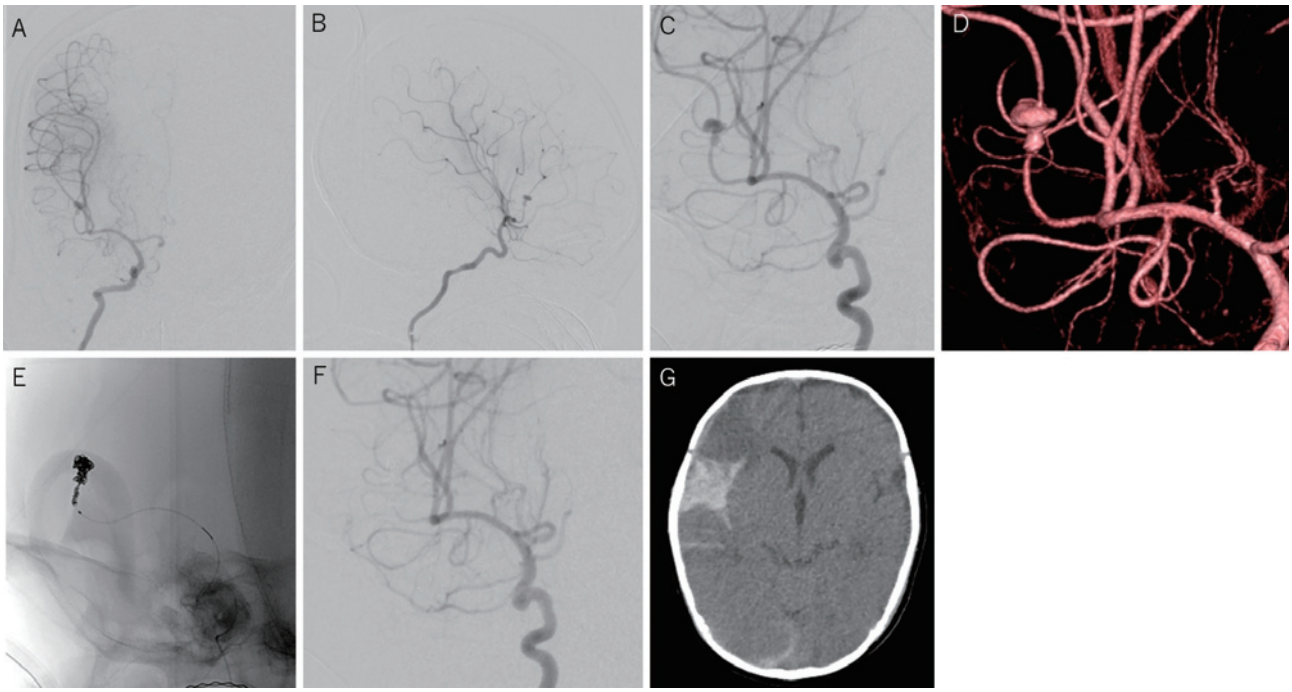

Fig. 2 (A) and (B) show preoperative cerebral angiography (frontal and lateral view). (C) and (D) show preoperative angiography and VRT-image (same frontal oblique angle). After coil embolization, the aneurysm and parent vessel were utterly occluded (E and F). CT scan on the day after surgery (G) shows a low-density area in the right frontal lobe consistent with an occlusion area.

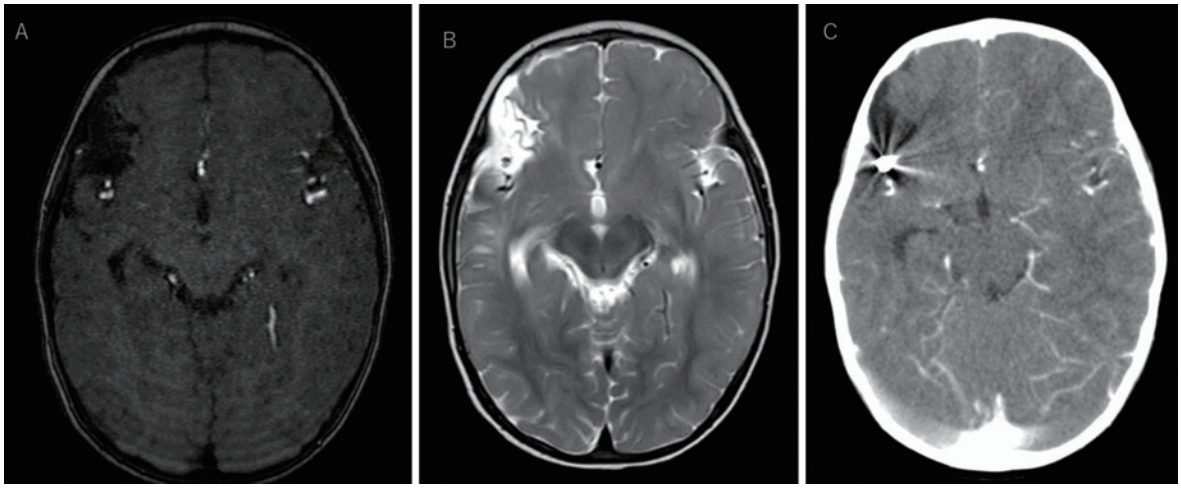

Fig. 3 Postoperative MRI after one year (A; MRA, B; T2-weighted image), the aneurysm, right M2 was not shown, and the cerebral tissue around the aneurysm had encephalomalacia. Postoperative 3D-CTA did not show after one year (C), the aneurysm, and right M2.

traumatic brain injury. Some dissecting aneurysms are related to medical genetic diseases such as, polycystic kidney disease, aortic coarctation, aortic valve stenosis, Moyamoya disease, sickle cell anemia, Kawasaki syndrome, Takayasu disease, tuberous sclerosis, neurofibromatosis type1, Ehlers-Danlos syndrome, or Marfan syndrome.[9] We diagnosed this case as a sporadic dissecting aneurysm because of the lack of IAs in both the parents and absence of signs of infection, connective tissue disease, or trauma. Owing to the limited experience with IAs in children, being a rarity, we conducted this literature review.

The PubMed database was used to perform an online search of reported studies using the following terms: "aneurysm," "subarachnoid hemorrhage," "subarachnoid bleeding," "infant," and "neonate." The number of cases was limited to less than 12 months. Only full-text articles published in English were selected. Papers were selected from the database inception until August 2022 to identify rele-

Table 1 Reported cases of subarachnoid hemorrhage under one year of age

Author and Year	Sex	Age (d; day, mo; month)	Presentation	Morphology	Location	Aneurysm size (mm)	Hemorrhage	Management	Outcome
Vapalahti PM et al. 1969[22]	F	3 mo	Coma	N/A	Rt. ACA	N/A	ICH	MS (Clipping)	Full recovery
Morelli RJ et al. 1977[23]	M	4 mo	Vomiting	Multilobulated	Lt. ACA	12	SAH	MS (Aneurysm resected)	Full recovery
Grode ML et al. 1978[24]	F	3 mo	Vomiting	N/A	Rt. MCA	N/A	SAH, ICH	MS (Clipping)	Full recovery
Roy C et al. 1990[25]	M	1.5 mo	Listlessness	N/A	Rt. MCA	N/A	SAH, ICH	MS (Clipping)	Death
DiMario FJ Jr et al. 1992[26]	F	10 mo	Seizure	Fusiform	BA	N/A	SAH, IVH	Autopsy	Death
Kuchelmeister K et al. 1993[27]	M	3 d	Seizure	Saccular	AcomA	10	SAH, IVH	Autopsy	Death
Tan MP et al. 1998[28]	M	11 d	Drowsiness, vomiting	Fusiform	ACA	10	SAH, IVH	MS (Clipping attempt)	Death
Plunkett J et al. 1999[29]	M	7 mo	Sudden death	Fusiform	BA	5	SAH	Autopsy	Death
Young WF et al. 2000[30]	M	1 mo	Apnea	N/A	Lt. MCA	N/A	SAH, ICH	MS (Coagulation of the neck)	Partial recovery
Jansen FE et al. 2000[31]	F	1 mo	Vomiting	N/A	Lt. BA-SCA	8	SAH, ICH	MS (Clipping)	Full recovery
Desai K et al. 2001[32]	F	4 mo	Sudden death	N/A	Lt. PICA	N/A	SAH, ICH	Autopsy	Death
Motohashi O et al. 2004[33]	F	1 mo	Seizure, vomiting	N/A	Rt. ACA	7	SAH	MS (Trapping)	Full recovery
Elgamal EA et al. 2004[34]	M	8 mo	Seizure	Saccular	AcomA	N/A	SAH	MS (Clipping)	Full recovery
Song JK et al. 2005[35]	F	11 d	Seizure	N/A	AcomA	N/A	SAH, IVH	EV (Coiling)	Full recovery
Bhardwaj G et al. 2010[36]	M	7 mo	Apnea	Fusiform	Rt. MCA	6	SAH, ICH, SDH	MS (Clipping)	N/A
Tai YP et al. 2010[37]	M	9 d	Poor activity	Fusiform	Rt. PICA	3	SAH, ICH, IVH	EV (Coiling + NBCA)	Full recovery
Ko A et al. 2010[38]	M	2 mo	N/A	Saccular	Lt. PCA	16	SAH, IVH	EV (Coiling)	Partial recovery
Mena OJ et al. 2011[39]	F	7 mo	Drowsiness	Fusiform	Lt. MCA	30	SAH	Autopsy	Death
Yatomi K et al. 2014[11]	F	1 mo	Seizure	N/A	AcomA	N/A	SAH	EV (Coiling)	Partial recovery
Su TM et al. 2014[40]	F	7 mo	Drowsiness, vomiting	Fusiform	Rt. PICA	N/A	SAH, IVH, SDH	Autopsy	Partial recovery
Thong KM et al. 2014[41]	M	6 d	Drowsiness, vomiting	Unknown	Unknown	N/A	SAH	Autopsy	Death
Fathi NQ et al. 2015[42]	F	2 mo	Seizure	N/A	Lt. ACA	31	SAH, ICH	MS (Trapping)	Partial recovery
Ravindra VM et al. 2016[43]	F	6 mo	Seizure	Saccular	Lt. MCA	6	SAH, SDH	EV (Onyx embolization)	Partial recovery

Table 1 Reported cases of subarachnoid hemorrhage under one year of age (continued)

Author and Year	Sex	Age (d; day, mo; month)	Presentation	Morphology	Location	Aneurysm size (mm)	Hemorrhage	Management	Outcome
Del Santo MA et al. 2016[44]	M	3 mo	Vomiting, drowsiness	Saccular	Rt. SCA	6.5	SAH	EV (Coiling)	Full recovery
Hidalgo J et al. 2017[45]	F	26 d	Seizure	Saccular	Lt. MCA	2	SAH, ICH, SDH	MS (Clipping)	Full recovery
Lyon KA et al. 2017[46]	F	1.5 mo	Seizures, drowsiness	Saccular	Rt. MCA	13	SAH	EV (Coiling)	Full recovery
Mohotti JE et al. 2018[47]	F	28 d	Seizure	Saccular	Rt. MCA	14	SAH, ICH	MS (Clipping)	Partial recovery
CreveCoeur TS et al. 2019[48]	M	4 d	Drowsiness, vomiting	Fusiform	Rt. AICA	1.6	SAH, IVH	EV (Onyx embolization)	Partial recovery
Lu VM et al. 2019[49]	F	7 mo	Seizure	Fusiform	Lt. MCA	20	SAH, SDH	MS (Trapping)	Partial recovery
Goia A et al. 2020[50]	M	21 d	Vomiting, seizure	Fusiform	Lt. MCA	7	SAH	EV (Coiling)	Full recovery
Saraf R et al. 2021[51]	F	11 mo	Anisocoria	Fusiform	Lt. MCA	N/A	SAH, ICH, IVH	EV (Coiling)	Full recovery
Barchetti G et al. 2021[52]	F	10 mo	Drowsiness	Fusiform	Rt. MCA	N/A	SAH, ICH	EV (FD)	Full recovery
Aldea CC et al. 2022[53]	M	4 mo	Drowsiness, vomiting	Saccular	AcomA	N/A	SAH, ICH	MS (Clipping)	Full recovery
Present Case 2022	M	3 mo	Seizures, drowsiness	Fusiform	Rt.MCA	13	SAH	EV (Trapping)	Full recovery

M; male, F; female, N/A; not available, MS; microsurgical, EV; endovascular, FD; flow diverter, ACA; anterior cerebral artery, MCA; middle cerebral artery, AcomA; anterior communicating artery, BA; basilar artery, SCA; superior cerebellar artery, SAH; subarachnoid hemorrhage, PICA; Posterior inferior cerebellar artery, ICH; intracerebral hemorrhage, IVH; intraventricular hemorrhage, SDH; subdural hemorrhage

vant articles. Only cases of spontaneous IA in infants, including infected IAs and traumatic IAs, were excluded. Table 1 summarizes the literature review that yielded 33 reported cases. The sex distribution was 15 boys and 18 girls, with seizure being the main clinical presentation (12 out of 33 patients). Subarachnoid hemorrhage in infants younger than three months has been reported in only 20 cases, making it extremely rare. In this review of less than 12 months, the main location was the MCA, accounting for 1/3 of the total (Table 1), but previous studies have shown that aneurysms in infants are more common in the anterior circulation (76%), especially in the MCA. In general, children have a high rate of dissecting aneurysms (16-45%), and more than half of the patients in this review also presented the fusiform type. It has been reported that the proportion of dissecting aneurysms increases at younger ages due to disruptions or defects in the vascular lamina due to infections or congenital diseases.[10,11] In a recent study focusing on the risk of seizure in pediatric patients with IAs, it appeared that pediatric patients are at a higher risk of seizure in the case of SAH as compared to the adults. The advanced hypothesis for this is the vulnerability of the cortex in pediatric patients, and the increased prevalence of IA located in the MCA is associated with cortical hemorrhage.[12,13] Of the 33 previously reported cases, a total of 5 cases underwent trapping, with 3 of these cases resulting in occlusion-territory infarction. The remaining cases lacked a comprehensive description of any associated complications.

Owing to the small number of cases, no algorithm has been established for treatment methods, and decisions must be made on a case-by-case basis. The following are some of the reasons why endovascular treatment is preferred: the difficulty of identifying the Sylvian fissure, distinguishing arteries from veins, the low volume of blood circulation, and the risk of anesthesia when compared to adults.[14,15] Furthermore, it is notable that children exhibit higher tolerance to vasospasm when compared to adults. This could be attributed to their augmented leptomeningeal circulation and cerebral blood flow.[16-19] On the other hand, recent advancements in surgical navigation systems and indocyanine green (ICG) video angiography have made it feasible to precisely locate blood vessels and aneurysms. As a result, when the parent vessel can be

maintained, microsurgical procedures like neck clipping should be considered a viable alternative. The aneurysm's morphology and perfusion area are key considerations in making such decisions.

The long-term outcomes of IAs in infants have been described recently in two studies. In 114 cases, with a mean follow-up of 25 years, long-term excess mortality was observed in pediatric patients presenting with IA compared to the general population. In the 1-year post-SAH survivors, the cause of death in 10% cases was SAH recurrence (10%) because of either de novo IA or its post-surgical recurrence. Long-term outcomes were favorable in 62% of the patients, whereas 35% succumbed to it, and 3% remained dependent. The factors associated with good outcomes were good clinical status at admission, IAs from the anterior circulation, and the absence of vasospasm. There is no difference in outcomes between microsurgery[20] and endovascular treatments, and similar results have been reported in the past too.[8,21]

Conclusion

IAs in infants are rare, and we report a case of a large ruptured dissecting aneurysm in the right MCA that was successfully treated in a 3-month-old infant with endovascular therapy. Since life expectancy is longer in children than in adults, the choice of therapeutic options is essential, and more such case reports are needed to decide the best treatment protocol.

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Informed Consent

The patient has consented to the submission of this case report to the journal.

Conflicts of Interest Disclosure

All authors declare that there are no conflicts of interest.

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