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Microsurgery for a ruptured intracranial aneurysm in a 3-year-old child: A case report

Hung Manh Ngo^a, Hung Thanh Chu^{b,*}, Dong Duc Nguyen^a

^a Department of Neurosurgery, Viet Duc Hospital, Hanoi, Viet Nam

^b Hanoi Medical University, No 1 Ton That Tung Street, Hanoi, Viet Nam

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ABSTRACT

INTRODUCTION: Pediatric intracranial aneurysms are rare, with some different characteristics from those in adults. Here, we present a case of distal anterior cerebral artery aneurysm which we believe the first case from Vietnam.

PRESENTATION OF CASE: A 3 years old boy presented with headache, lethargic and hemiparesis was diagnosed ruptured distal anterior cerebral artery aneurysm with CTA. 10 days before admission in our hospital, the toddler was admitted in local hospital with diagnosis of SAH without more accurate findings. Clipping microsurgery of aneurysms with left interhemispheric approach was done with good outcome.

DISCUSSION: The epidemiology, presentation and diagnosis and strategy of treatments as well as their outcomes were discussed. We also discussed our thoughts about our case within the limited-resource condition such as in Vietnam.

CONCLUSION: Pediatric intracranial aneurysms are rare but should be recognised in neurosurgical practice. Surgery is an effective treatment method.

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1. Introduction

Intracranial aneurysm (IA) is a common cerebrovascular disease in adults but not common in children. Pediatric intracranial aneurysms accounts for 0.5–4.6 % of all aneurysms [1].

Intracranial aneurysms in pediatric population differ from those in adults in their various and peripheral location [2]. Pediatric patients with IAs have subarachnoidal hemorrhage (SAH) as the most popular clinical presentation, with good clinical grade (Hunt-Hess). Therefore, IAs are often ignored and diagnosed belatedly [3]. IAs in children are unrecognised in developing countries such as Vietnam, where relevant radiology imaging machines, knowledge and experiences remain insufficient.

In this paper, we reported a ruptured intracranial aneurysm in a 3-year-old patient. We believe this is the first case report on this

issue in Vietnam. This case report has been reported in line with the SCARE criteria. [4]

2. Case presentation

A 3-year-old boy with no medical history complained of mild to moderate headache, headache and vomiting. The patient was brought to local hospital. On examination, he was alert, had no fever and no paralysis. Non-enhanced computed tomography (CT) scan showed hemorrhage in frontal interhemispheric fissure (Fig. 1A). After 5 days of conservative treatment, the patient was discharged.

5 days later, the patient went to local hospital again, complained of severe headache, lethargic, and right hemiparesis. He had a brain CT scan showing subdural and intracerebral hematoma in the frontal lobe with intraventricular hemorrhage (Fig. 1B), then he was transferred to our hospital.

On examination, he was lethargic and suffered right hemiparesis with muscle strength of 3/5. The CT angiography showed an aneurysm at the distal anterior cerebral artery (ACA), with the dimension of 12 × 7 mm and the neck of 2 mm. (Figs. 1C and 2 A, B). The Hunt-Hess grade was 3, mFisher score was 4, modified Rankin Scale (mRS) was 4.

In our hospital, it was unable to take the Digital Subtraction Angiography (DSA) as well as endovascular treatment in emergency setting. The surgical decision was made after discussion with his parents. The surgery was made by a senior doctor.

Abbreviations: DSA, digital subtraction angiography; CT, computed tomography; CTA, computer tomography angiography; ACA, anterior cerebral artery; A2, A2 segment of anterior cerebral artery; ICA, internal carotid artery; AN, aneurysm; IA, intracranial aneurysm; SAH, subarachnoidal hemorrhage; IVH, intraventricular hemorrhage; EVD, external ventricular drain; ICU, Intensive care unit; mRS, modified Rankin Scale; GOS, Glasgow Outcome Score; LOC, Loss of consciousness.

* Corresponding author.

E-mail addresses: ngomanhhung2000@gmail.com (H.M. Ngo), hungchuthanh@gmail.com (H.T. Chu), nguyenducdong.2293@gmail.com (D.D. Nguyen).

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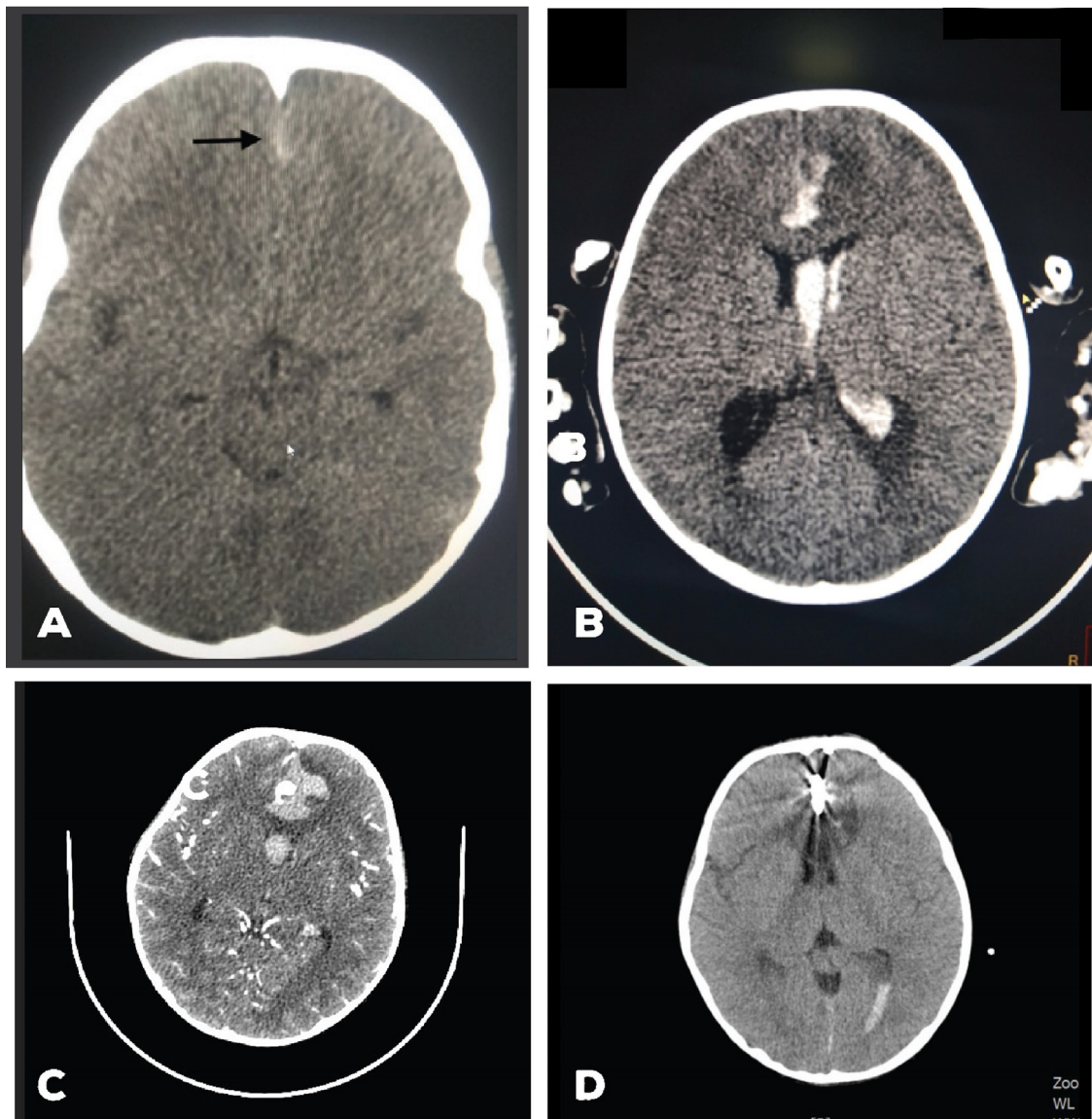


Fig. 1. A: The first non-enhanced CT scan showed interhemisphere subdural hemorrhage.
 B: The second non-enhanced CT scan showed interhemisphere subdural and intracerebral hemorrhage, with intraventricular hemorrhage.
 C: The enhanced CT scan showed interhemisphere subdural and intracerebral hemorrhage, with intraventricular hemorrhage due to rupture of distal ACA.
 D: The postoperative CT scan showed no subdural or subarachnoid hemorrhage, no hydrocephalus.

Under general anesthesia, the patient's position was supine with slight elevated-straight head fixed with Mayfield clamp. Left frontal extended over superior sagittal sinus craniotomy was taken.

The dura matter was incised and finished at the left border of superior sagittal sinus. The interhemispheric fissure was exposed without cutting the falx. We continued dissecting proximal pericallosal artery aneurysms (A2 segment of the anterior cerebral artery) and found the aneurysm at the bifurcation between the distal ACA and the frontopolar branches of A2. Two mini-clips were placed on the neck of the aneurysm (Fig. 3).

Intracerebral hematoma was then evacuated and hematoma in the left lateral ventricular was aspirated with irrigation. After that, we performed external ventricular drain (EVD) in the left lateral ventricular. EVD was then closed.

After surgery, the toddler was transferred to Intensive care unit (ICU) for 2 days and the patient was administered intravenous

nimodipine with the dose of 1 mg/kg q4h for 5 days, followed by oral nimodipine for 15 days, as in Heffren's regimen [5].

Two days after the operation, the EVD was rejected after brain CT scan showed neither hydrocephalus nor intraventricular hemorrhage (IVH) (Fig. 1D). Nine days after, computer tomography angiography (CTA) was done, showing no residual aneurysm, and no vasospasm (Fig. 4A,B). He was then discharged, with mRS of 2, improved hemiparesis with muscle strength of 4/5. At one-month postoperative follow-up, mRS was 1, the patient complained of some mild headache with a complete recovery from hemiparesis.

We planed a reexamination and CT-scan in 3 month. Due to the epidemic of Covid-19, the patient was unable to come to the follow-up. By phone, we figured out the patient's Glassgow Outcome Score (GOS) 5 and mRS 0. The children no longer suffered from aphasia, hemiparesis and headache.

At one-year follow-up, mRS was 0, GOS was 5, CTA showing no residual or recurrent aneurysm, no hydrocephalus (Fig. 4C, D).

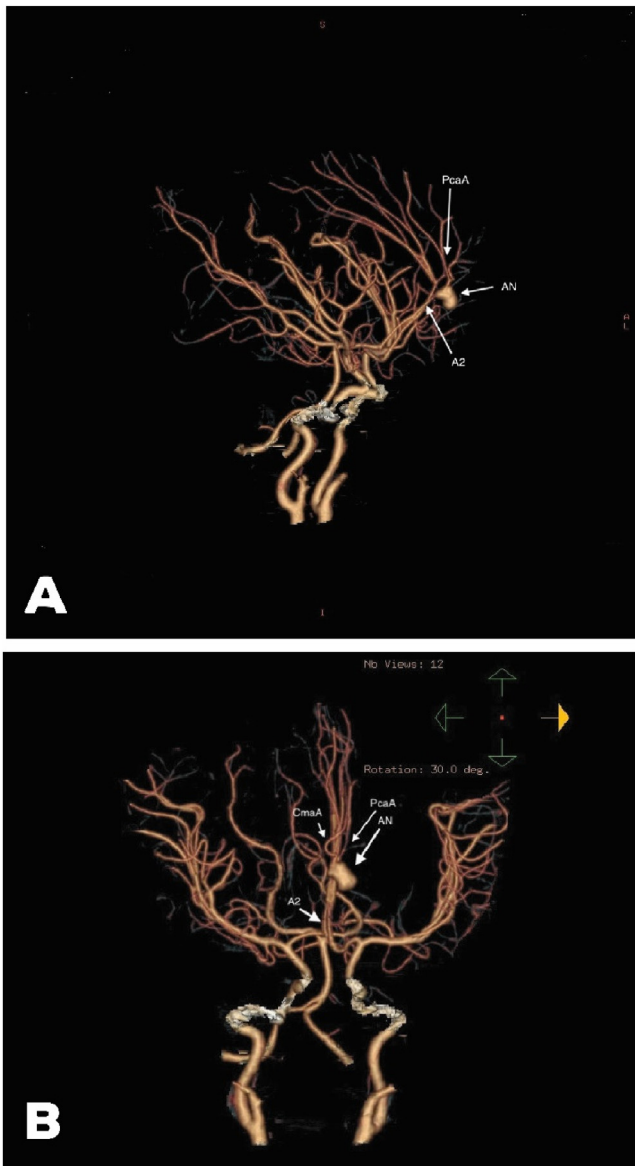


Fig. 2. A,B: The CT angiography showed an aneurysm in distal ACA. PcaA: Pericallosal artery. CmaA: Callosal marginal artery. AN: Aneurysm. A2: A2 segment of ACA.

3. Discussion

The incidence of intracranial aneurysms (IAs) in pediatric population is rare, ranging from 0.5 to 4.6% [6]. 75 % of them located in anterior circulation, while 25 % in the posterior circulation [7]. The most common location was the internal carotid artery (ICA), accounting for 27 %, followed by the middle cerebral artery (26 %) [7]. Rarely, the distal of ACA, such as pericallosal (5.4 %) [3], and distal ACA (2 %–4.11 %) [7,8]. Multiple aneurysms were encountered in 8.7 % of 50 patients in 22 cohort studies [7]. The mean age at diagnosis varied from 7.6–14.3 [3–8]. Of them, 66 % was male [5].

The most common clinical presentation was headache (61 %–75 %) [8,9], followed by Loss of consciousness (LOC) (49 %); seizures (35 %), and limb weak (16 %) [8].

The incidence of ruptured IAs at time of diagnosis range from 55 %–87 % [7–11], while 26 % of patients suffered from rebleeding before diagnosis/ treatment [3]

In pediatric population, intracranial aneurysms are associated with some well known diseases such as sickle cell disease (SCD), Marfan syndrome. However, the rate of this comorbidity was

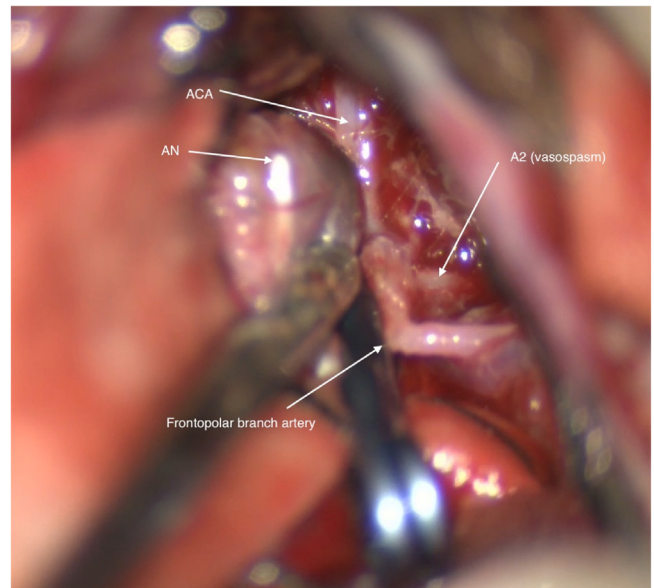


Fig. 3. The intraoperative image showed aneurysm in distal anterior cerebral artery. ACA: Anterior cerebral artery. AN: Aneurysm. A2: A2 segment of anterior cerebral artery.

unpopular. Koroknay-Pal et al. reported that 7 children in 114 cases had risk factors associated with intracranial aneurysms [3]. This rate was varied, as 0 in 22 case [11] or 27 % [9]. Lasjaunias et al. reported that in 59 cases of patients under 15 year olds, 5 of them had familial diseases [12]

Treatment for intracranial aneurysms aims to exclude the aneurysms from the blood stream without disrupting the normal blood supply. Treatment options for ruptured IAs include microsurgery and endovascular coiling, while conservative treatment is acceptable for unruptured cases. For children, life span expected for them is a factor supporting an enduring treatment.

Endovascular treatment had been utilized widely in pediatric intracranial treatment. The rate of utilizing endovascular treatment compared to surgery from 2000 to 2015 has increased as reported by Beez et al. Yasin et al. (2019) reported that in 57 cases, endovascular treatment rate was 44 %, surgery was 23 % (the other 33 % of cases patients received conservative treatment [9]. In a meta-analysis report with 560 cases of pediatric intracranial aneurysm, Yasin et al. figured out that there was no significant difference in long-term result of these two treatment methods [9].

Our patient was treated by surgery for the following reasons. We were unable to performed DSA and endovascular treatment in our emergency setting. However, intracranial aneurysm surgery was a familiar treatment in our center. As the intracranial aneurysm ruptured, the hematoma caused neurological deterioration and hemiparesis, while intraventricular hemorrhage could lead to hydrocephalus. The hematoma mass effect and risk of hydrocephalus favored the use of surgery over endovascular treatment.

After evacuation of the hematoma, we performed EVD in the left lateral ventricular and closed the drain. In our center, it is often strenuous to start emergency surgery. EVD appeared useful in prevention of postoperative hydrocephalus, as the drain would be opened instantly when the patient had signs of deteriorated neurologic functions and evidence of hydrocephalus.

The recurrence risk of ICA in pediatric population was higher than that in adult’s one [3,5]. Endovascular treatment was supposed to be more related to the recurrence risk than surgery, as it required longer term of follow-up, and endovascular materials were perceived as non-durable. Besides, the cost of treatment was

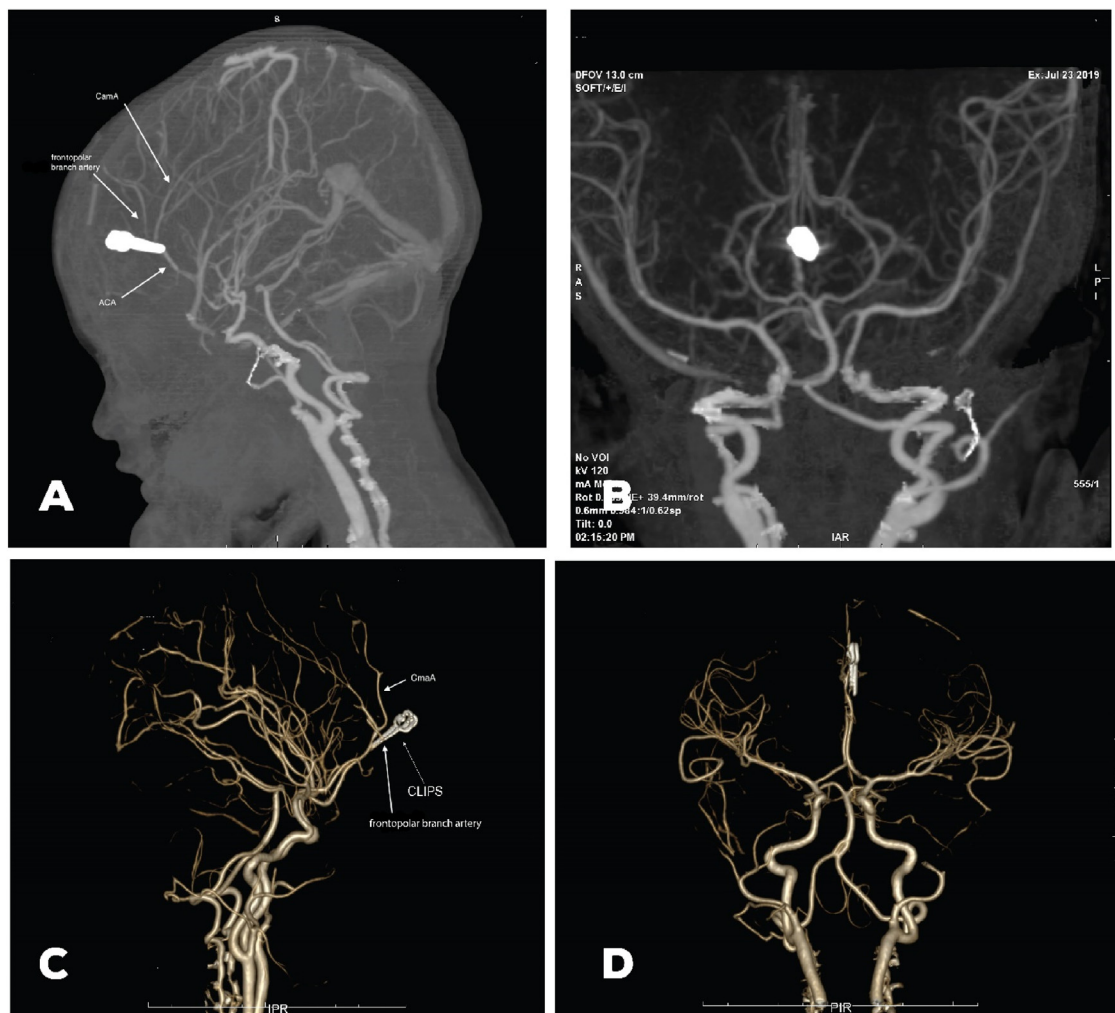


Fig. 4. The postoperative CT angiography showed clipping of aneurysm in distal cerebral artery. CmaA: Callosalmarginal artery.

one essential factor supporting our decision, since endovascular method costs much higher than surgery in Vietnam [13].

Cerebral vasospasm was one of the complications causing disability in SAH after ruptured ICA. The risk of cerebral vasospasm in pediatric ruptured ICA varied from 9.4%–14.3% [4,11] to 23% [5], lowered than in adult (81%), with less severe symptoms. Our center used the same treatment regimen for cerebral vasospasm as Heffren et al. [2]. The hemorrhage and vasospasm after ICA in pediatric population appeared to cause less severe consequences than in adults, owing to the leptomeningeal collateral diversity in children [3].

4. Conclusion

Pediatric intracranial aneurysms are rare but should be recognised in neurosurgical practice. Surgery is an effective treatment method.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

The study was approved by the Research Ethics Committee of Hanoi Medical University. The procedures used in this study adhere to the tenets of the Declarations of Helsinki.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author's contribution

Hung Manh NGO: the main doctor conceived the original idea and operated the patients, revised manuscript.

Hung Thanh CHU: followed up, wrote manuscript.

Dong Duc NGUYEN: data curation, followed up, revised manuscript.

Guarantor

Hung Manh NGO, MD, PhD.

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Availability of data and material

Data is available upon reasonable request and with permission of Viet Duc Hospital. No patient or author details are included in the figures.

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