

# The clinical features and treatment of pediatric intracranial aneurysm

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Received: 20 August 2008 / Published online: 7 October 2008  
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## Abstract

**Purpose** Intracranial aneurysms are relatively rare in the pediatric population. The objective of this study was to highlight the clinical and radiological features and the therapeutic outcome and clarify the choice of therapeutic strategies for pediatric intracranial aneurysms.

**Materials and methods** Twenty-four consecutive children (age  $\leq 14$  years) who were diagnosed and treated for intracranial aneurysms in our institute in the last 23 years were included in this study.

**Results** There were nine (36%) patients with posterior circulation aneurysms and eight (32%) with giant aneurysms. Eleven (46%) patients presented with subarachnoid hemorrhage. Fifteen patients underwent endovascular treatment, and four received microsurgical therapy. Five patients were treated conservatively. Ninety-two percent ( $n=22$ ) of the patients showed favorable outcomes.

**Conclusions** Pediatric intracranial aneurysms differ in many ways from those in adults: male predominance; high incidence of giant, dissecting, and fusiform aneurysms;

high incidence of aneurysms in the posterior circulation; high incidence of spontaneous thrombosis; better Hunt–Hess grades at presentation; and better therapeutic outcome. For children with intracranial aneurysms, both microsurgical approaches and endovascular treatment were effective. For many complex aneurysms, endovascular therapy was the best choice.

**Keywords** Pediatric · Aneurysm · Intracranial · Outcome · Therapy

## Introduction

Intracranial aneurysms are extremely uncommon in the pediatric population (age  $\leq 14$  years), with a reported prevalence ranging from 0.5% to 4.6% [1–3]. Their etiology is poorly understood, and certain features such as the location, morphology, presentation, and therapeutic outcome make them unique in comparison with intracranial aneurysms in adults. To date, we do not have a great enough number of cases to sufficiently provide a meaningful characterization of this disease [4]. In this study, we have tried to analyze data from pediatric intracranial aneurysm patients in order to provide a better insight into the epidemiology, clinical and radiological features, outcome, and choice of therapeutic strategies.

## Materials and methods

Twenty-four consecutive children (age  $\leq 14$  years; 14 boys, ten girls; male/female ratio=1.4:1; mean age  $8.83 \pm 2.7$  years, age range 1–14 years) who were diagnosed and treated for intracranial aneurysms at our institute between

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October 1985 and July 2008 were included in this study. We retrospectively collected all the hospital records and follow-up data of these patients from the aneurysmal database at the Cerebrovascular Diseases Research Institute, Beijing, China.

At admission and before surgery, all patients were graded according to Hunt–Hess classification. Grades 0–3 were considered as good preoperative status, and grades 4 and 5 were considered as poor status. Patients without subarachnoid hemorrhage (SAH) history were included in grade 0. Besides Hunt–Hess classification, all the patients were graded according to the Fisher grading system.

For patients whose aneurysms were treated by a microsurgical technique, we performed digital subtraction angiography (DSA) before discharge to check whether there were residual aneurysms.

Twenty-one patients were followed up postoperatively at 3, 6, and 12 months and subsequently at yearly intervals; DSA was performed in most patients at 3 or 6 months to check if aneurysms had recurred. The mean follow-up was  $13.6 \pm 9.2$  months (range 1–32 months). On discharge and every follow-up, outcome was assessed using the Glasgow Outcome Scale. A score of 5 or 4 was taken as a favorable outcome, whereas scores of 3, 2, or 1 were considered as unfavorable.

## Results

### Preoperative state

According to Hunt–Hess classification, 21 patients were in a good preoperative state: 13 were in grade 0, six in grade 2, and two in grade 3. Three patients in grade 4 were in a poor preoperative state.

According to the Fisher grading system, 13 patients were in grade 1 (no history of SAH), five were in grade 2, two were in grade 3, and four were in grade 4.

### Clinical presentation

The predominant clinical presentation of patients in our series included the following: sudden severe headache in 11 patients due to SAH, chronic or intermittent headache in five patients, dizziness in four patients, proptosis and diplopia in three patients, hemiparesis in two patients, and loss of consciousness in one patient.

### Radiological characteristics

All patients underwent DSA on admission to clarify the location, size, shape, and number of aneurysms. In our series, there was a total of 25 aneurysms in 24 patients, with

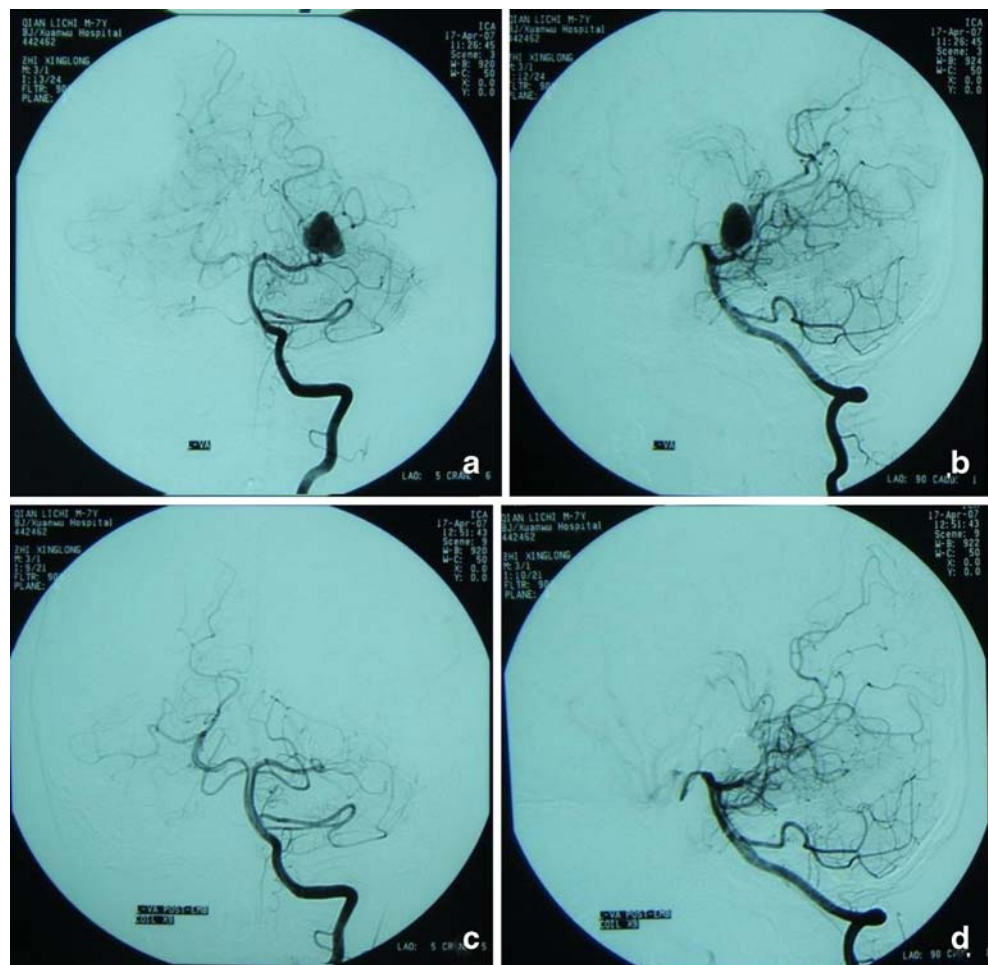
one patient having two aneurysms. On angiography, 18 aneurysms were of the saccular type, and seven were of the fusiform or irregular type. One aneurysm was mycotic. There were six small, 11 large, and eight giant aneurysms. Nine aneurysms were located in the posterior circulation and 16 in the anterior circulation. The internal carotid artery (ICA) was the most common site for aneurysms ( $n=7$ ). Three aneurysms were located in the cavernous segment of the ICA. Two aneurysms were located in the ICA bifurcation, and the remaining two aneurysms were located in the supraclinoidal ICA and origin of the posterior communicating artery, respectively. The middle cerebral artery (MCA) was next most common site for aneurysms ( $n=5$ ). Three aneurysms were located in the MCA bifurcation and two in the distal MCA territory. As for the anterior cerebral artery, two aneurysms were located in the anterior communicating artery, and two were located in the distal anterior cerebral artery (A2 segment). In our case series, there was only one patient who had multiple aneurysms. The sites of aneurysms that were located in the posterior circulation included the basilar artery ( $n=3$ ), vertebral artery ( $n=2$ ), and posterior cerebral artery ( $n=4$ ). One patient had a medical history of bacterial endocarditis and atrial septal defect, and another had a history of head trauma (Figs. 1, 2, and 3; Tables 1).

### Therapeutic outcome

Fifteen patients underwent endovascular therapy. Four patients were treated by using a microsurgical technique, three by aneurysmal neck clipping, and one by parent artery ligation because of difficulty in dissection of the aneurysmal neck. Five patients (six aneurysms) received conservative therapy. The parent artery and aneurysm regressed spontaneously prior to treatment in two patients. Two patients received conservative therapy due to a good clinical state on admission and fear of operative risk. Another patient with a giant dissecting aneurysm in the right vertebral artery preferred further follow-up and refused surgery because of the doubled operative risk and economic reasons.

There were 22 patients who had a favorable outcome and two patients who had an unfavorable outcome. There was one death in the series. This patient (No. 12), who was in good grade at admission, had a giant irregular dissecting aneurysm in the basilar artery trunk. The right vertebral artery had undergone hypoplasia and did not connect with the basilar artery. During surgery, we occluded the left vertebral artery distal to the origin of the posterior inferior cerebellar artery so as to trap the aneurysm. During the interventional therapy, the patient was stable, but after the operation, the condition of this patient gradually worsened, and the patient finally died due to occlusion of the perforating branch to the brainstem. Another patient with

**Fig. 1** Patient no. 8 who was a 7-year-old boy with a left P2 saccular aneurysm (19 mm×17 mm). Digital subtraction angiography images: **a** anterior–posterior view before surgery, **b** lateral view before surgery, **c** anterior–posterior view after surgery, **d** lateral view after surgery



poor outcome was a 7-year-old boy. She had a left middle cerebral artery aneurysm. The aneurysm was complex (both M2 segments originated from the aneurysm). We planned to trap the aneurysm after the superior temporal artery to MCA bypass. However, the aneurysm ruptured again preoperatively, and she was in a deep coma state and had a brain hernia, so we had to occlude the parent artery and the aneurysm in such an emergent state. On discharge, this patient had right hemiparesis and partial dysphasia.

## Discussion

### Epidemiology

The incidence of pediatric intracranial aneurysms was very low. According to previous reports, the prevalence of pediatric aneurysms among all intracranial aneurysms has ranged from 0.5% to 4.6% [1–3]. In our institute, we diagnosed and treated 24 pediatric aneurysmal patients during the past 23 years, nearly one patient per year. During the same period, we diagnosed and treated a total of 1,750 intracranial aneurysm cases, and therefore, the incidence of

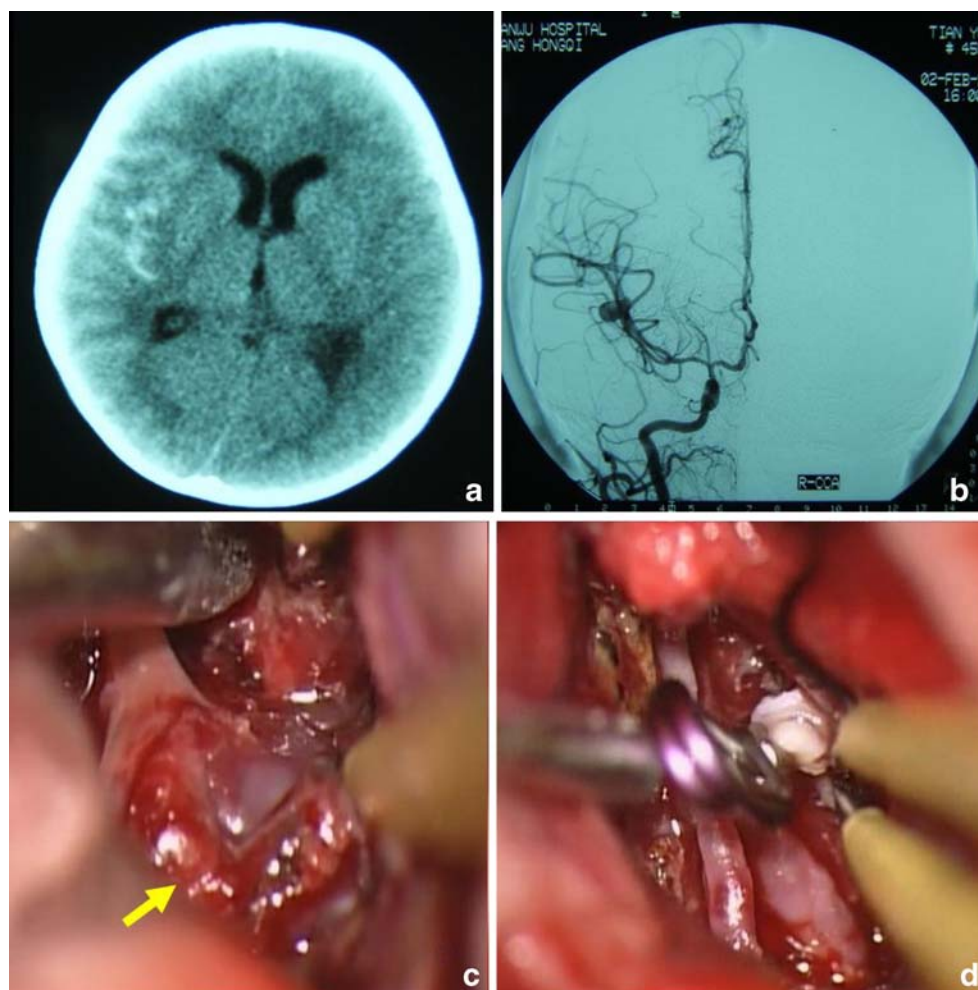
pediatric aneurysms was 1.37% (24/1750), which is similar to previous reports.

In adults, women are more likely to have intracranial aneurysms than men. Women have between three to five times more aneurysms than men [5]. However, in children, there is no predilection for female patients. On the contrary, in the pediatric literature, intracranial aneurysms have occurred more often in boys than in girls [6]. This male dominance was also found in our study. The ratio between males and females was 1.4:1.

### Etiology and pathogenesis

The etiology and pathogenesis of pediatric intracranial aneurysms are still unknown. Allison et al. regarded it as a congenital disease [7]. Agid et al. believed that neither adult nor pediatric aneurysms were truly congenital in nature [8]. Other authors reported that the occurrence of an aneurysm was probably the result of an interplay between structural changes in the vessel wall and hemodynamic stress [9, 10]. Possible mechanisms for the development of aneurysms in children were postulated by Lasjaunias et al. [6]. They suggested that aneurysms in children must be the expression

**Fig. 2** Patient no. 3 who was a 2.5-year-old girl. **a** CT scan showed subarachnoid hemorrhage in the right sylvian fissure; **b** angiography showed an aneurysm in the right MCA distal branch; **c** an aneurysm located in the distal branch of right MCA (*arrow*) during the operation; **d** clipping the aneurysm and cutting the body of the aneurysm



of various vessel wall dysfunctions, which result in transient or permanent failure to repair a partial insult. In addition, associated conditions, i.e., congenital, traumatic, and infectious diseases (Marfan's syndrome, Ehler–Danlos syndrome, polycystic kidney disease, pseudoxanthoma elasticum, sickle cell anemia, tuberous sclerosis, bacterial endocarditis, closed and penetrating head injury, irradiation) also contribute to aneurysms in the pediatric population. In our study, one (4%) aneurysm was secondary to trauma, and one (4%) infectious aneurysm was due to bacterial endocarditis.

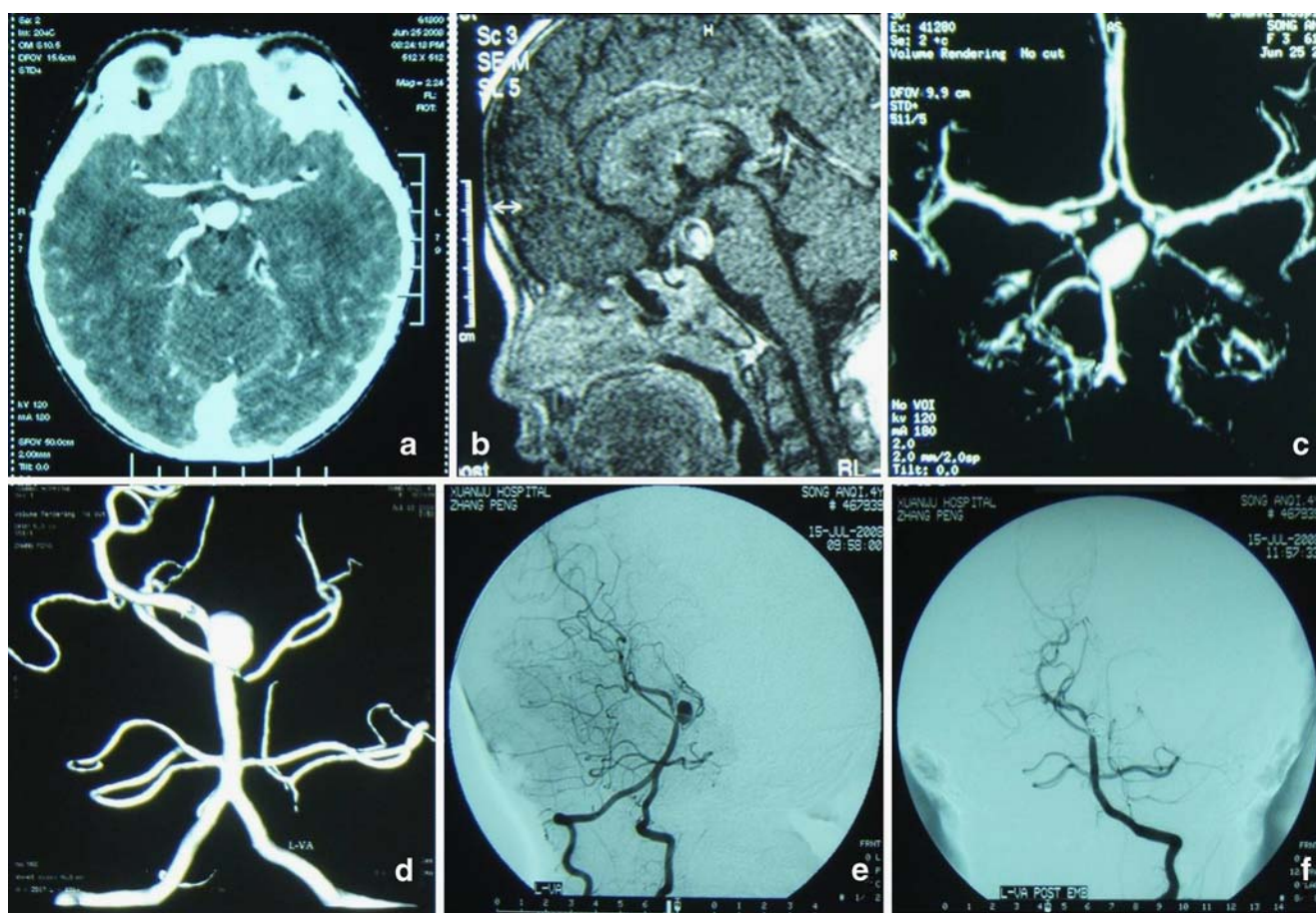
#### Presenting features

The presenting features of intracranial aneurysms in the pediatric population are different from those in adults. The incidence of SAH in previous reports has varied from 35% to 100% [11–13]. Sharma et al. reported that the incidence of SAH was 82% in their case series, and the majority (86%) of those who presented with SAH had good Hunt–Hess grades [14]. In our case series, the most common presentations were acute severe headache caused by SAH. The incidence of SAH was 46%, which was lower than in

the above reports. Besides SAH, the presentations in this group were focal neurological symptoms from mass effect such as proptosis, diplopia, hemiparesis, chronic or intermittent headache, dizziness, and so on. The incidence of mass effect was remarkably higher (54%) than in other reports [11, 12]. Sharma et al. reported that the incidence of seizures was 18.2% [14]. However, in the present series, there were no patients who presented with seizure.

#### Morphological characteristics

**Location** The location of the aneurysms is also different in children. According to most literature, the commonest site for pediatric intracranial aneurysms is the ICA bifurcation, with the incidence ranging from 24% to 50% [4, 13, 15–17]. However, we did not find this predilection for ICA bifurcation site in our case series. Only three (12%) aneurysms occurred at the ICA bifurcation. This was inconsistent with reports by other authors [11, 18]. There were three aneurysms located in the MCA bifurcation, three in the cavernous segment of the ICA, three in the basilar artery, and three in the posterior cerebral artery. According



**Fig. 3** Patient no. 4 who was a 3.5-year-old girl with a basilar artery bifurcation aneurysm. **a** CT enhanced image showed a lesion in the sellar region; **b** lateral view of vascular lesion in front of midbrain; **c** computed tomography angiography showed the aneurysm at the

basilar tip; **d** 3-D image showed the aneurysm; **e** oblique view showed the aneurysm pre-embolization; **f** anterior–posterior view before surgery

to the literature, the higher incidence of posterior circulation aneurysms is another characteristic of the pediatric population [7]. In our study, 36% (9/25) of the aneurysms were located in the posterior circulation.

**Size** Large and giant aneurysms are very common in the pediatric population compared with adults [6]. The reported incidence in this age group is from 20% to 45% [19]. There were 11 large, eight giant, and only six small aneurysms in our study. We did not find any small aneurysms in the posterior circulation. The incidence of giant aneurysms was 32% (8/25), which was in line with the reported incidence in the literature [11].

**Dissecting aneurysm** Nontraumatic dissecting aneurysms were usually located at the level of the supraclinoid ICA, the MCA, and the posterior circulation, especially at the P1 and P2 segments of the posterior cerebral artery [20]. The diagnostic standard for dissection in angiographic appearance includes preaneurysmal narrowing and fusiform shape [8]. Most of the dissecting aneurysms in our series were

located in the posterior circulation (five out of eight). Our series contained more nontraumatic dissecting aneurysms (eight out of 25, 32%) than other reports [21].

**Multiple aneurysms** The incidence of multiple aneurysms in children has been reported to be considerably lower than that in adults [22]. In our study group, only one patient (1/24) had multiple aneurysms. This patient had two aneurysms, which were located at the anterior communicating artery and left pericallosal artery, respectively.

**Spontaneous thrombosis**

In our case series, there were two patients whose aneurysms and parent artery had complete spontaneous thrombosis before treatment. The incidence was 2/24 (8.3%). Some previous reports have recorded this phenomenon [23]. In a report by Lasjaunias, ten patients had complete or partial spontaneous thrombosis among a total of 59 pediatric intracranial aneurysm patients. Unfortunately, the exact

**Table 1** Clinical characteristics of 24 children with 25 intracranial aneurysms

Case no.	Sex	Age (years)	No. of bleeds	Fisher HHS	Site	Size	Therapy	Glasgow Outcome Scale (GOS)
Patients who underwent surgery					HHS <sup>1</sup>			
1	M	1	1	3	3 L-M3	Large	Parent artery occlusion	5
2	M	2	None	1	0 R-MCA bifurcation	Large	Clip	4
3	F	2.5	1	2	2 R-M3	Large	Clip	5
4	F	3.5	None	1	0 BA-bifurcation	Large	GDC coiling	5
5	F	4	None	1	0 L-P2	Large	Parent artery occlusion	5
6	F	7	None	1	0 L-intracavernous ICA	Large	Parent artery occlusion	5
7	F	7	2	4	4 L-MCA bifurcation	Small	GDC coiling	3
8	M	7	None	1	0 L-P2	Large	GDC coiling	5
9	M	8	1	2	2 L-MCA bifurcation	Giant	Clip	4
10	M	9	None	1	0 R-intracavernous ICA	Giant	Parent artery occlusion	5
11	M	10	None	1	0 Basilar trunk	Giant	Parent artery occlusion	5
12	M	12	None	1	0 Basilar trunk	Giant	Parent artery occlusion	1
13	M	12	1	2	2 L-PcoA	Small	Clip	5
14	F	12	None	1	0 R-intracavernous ICA	Giant	Parent artery occlusion	5
15	M	13	1	3	3 L-vertebral artery	Large	GDC coiling	4
16	F	13	1	2	2 R-P3-4	Large	GDC coiling	5
17	F	14	1	4	2 L-ICA bifurcation	Small	GDC coiling	5
18	M	14	None	1	0 R-ICA superclinoid	Giant	GDC coiling+parent artery occlusion	5
19	M	14	None	1	0 R-PCA-BA	Giant	Stent	5
Patients who did not undergo surgery					HHS <sup>2</sup>			
20	M	5	1	4	2 ACoA; L-A2	Small; large	Conservative therapy	4
21	F	9	None	1	0 R-ICA bifurcation	Large	Spontaneously thrombosis	5
22	M	9	None	1	0 R-vertebral artery	Giant	Conservative therapy	5
23	M	11	2	4	4 ACoA	Small	Spontaneously thrombosis	4
24	F	13	1	2	2 ACoA	Small	Conservative therapy	5

Small diameter ( $\Phi$ ) < 10 mm, large 10 mm <  $\Phi$  < 25 mm, giant  $\Phi$  > 25 mm, L left, R right, HH Modified Hunt–Hess grading, GOS Glasgow Outcome Scale, ICA internal carotid artery, A2 A2 segment of the anterior cerebral artery, ACoA anterior communicating artery, MCA middle cerebral artery, M3 M3 segment of middle cerebral artery, PCoA posterior cerebral artery, PCA posterior cerebral artery, BA basilar artery, P2 P2 segment of posterior cerebral artery, P3-4 P3–P4 segment of posterior cerebral artery

<sup>1</sup> Hunt-Hess Scale before surgery

<sup>2</sup> Hunt-Hess Scale on admission

mechanisms and factors responsible for this occurrence cannot be predicted with sufficient reliability.

### Treatment

Just as for aneurysms in adults, the treatment for pediatric intracranial aneurysms has undergone significant evolution in recent years. At presentation, multimodal treatment strategies are now widely used as a standard approach and have therefore resulted in significant improvements in patient outcomes [14, 24].

The vast majority of patients, 87% (13 out of 15), who underwent endovascular treatment had a good recovery; only 13% (two out of 15) had a worse outcome. One boy with basilar trunk giant dissecting aneurysm died after surgery involving the left vertebral artery owing to thrombosis in the basilar artery. Therefore, the mortality rate was 4.2% in our series.

The choice of treatment method for a given aneurysm in our institution is made by a multidisciplinary team of neurosurgeons and interventional neuroradiologists. The endovascular approach is preferred whenever possible technically and is less dependent on the patient's clinical situation. We expect that with the evolution of intracranial stent technology, it is likely that in the future, less deconstructive endovascular procedures will be performed, and parent vessel preservation will be achieved more often.

Endovascular treatment of aneurysms in children is following the path of endovascular treatment of vascular malformations in children and of aneurysms in adults. With regard to treating aneurysms, the goal should be not only to achieve alleviation of the acute symptoms (such as due to mass effect) but also, most importantly, to protect the patient from a future bleed. The endovascular approach can only be considered if it achieves both these goals. Most of the literature regarding results of endovascular treatment for

pediatric aneurysms consists of case reports [25]; there have been only a few published reports on outcome in larger series [13]. Most of the case reports present a good outcome and successful occlusion of the aneurysm, with either reconstructive or deconstructive methods [17, 18, 20, 21]. Lasjaunias et al. reported a similar distribution among endovascular, surgical, and conservative approaches in their series as we report here [6]. They treated eight patients out of 20 (40%) by endovascular means and were successful in seven. These included four parent vessel occlusions and two cases of aneurysm coil embolization with preservation of the parent artery. In one case, endovascular treatment failed, and the patient received surgical treatment. All of the seven successful endovascular procedures resulted in complete regression of symptoms. Three of their patients were treated by surgery (15%; two with success and one with failure), seven patients (35%) were treated conservatively, and two (10%) had spontaneous thrombosis of the aneurysm. In the series of Proust et al., among 22 children, a microsurgical procedure was performed in 17 (77.3%), the endovascular approach was used in four (18.2%), and a combined approach in one (4.5%) [13]. The overall outcome was favorable in 63.6%; the mortality rate was 22.7%.

The reason why we chose the endovascular method to treat most of the pediatric patients with intracranial aneurysms in our case series mainly included two considerations: On one hand, the majority of aneurysms in our series were large or giant and more than one third of the aneurysms located in the posterior circulation. These radiological characteristics meant that there would be great difficulties if we choose a microsurgical method. Also, after many years of treating adults with intracranial aneurysms, we have become familiar with interventional skills and accumulated much experience for treatment of pediatric aneurysms.

With regard to a good clinical outcome in our study as opposed to poorer outcome in previous reports, we think that the development of endovascular or microsurgical technique skills has produced better results. Another important factor for good outcome in our series was that the majority of pediatric patients were in a good state on admission.

## Conclusions

Pediatric intracranial aneurysms have many clinical and radiological characteristics that differ from intracranial aneurysms in adults: male predominance; high incidence of giant, traumatic, infectious, dissecting, and fusiform aneurysms; high incidence of aneurysms in the posterior

circulation; high incidence of spontaneous thrombosis; and better Hunt–Hess grades at presentation. Pediatric intracranial aneurysms also have a better therapeutic outcome. For intracranial aneurysms in children, both microsurgical approaches and endovascular treatment were effective. For many complex pediatric intracranial aneurysms, endovascular therapy was the best choice.

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