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Intracranial aneurysms in adolescents

Jian-tao Liang • Li-rong Huo • Yu-hai Bao • Hong-qi Zhang • Zhen-yu Wang • Feng Ling

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Abstract

Purpose Intracranial aneurysms are extremely uncommon in adolescents. This study was undertaken to assess the clinical and radiological characteristics and clarify the choice of therapeutic strategies of intracranial aneurysms in adolescents with age range from 15 to 18 years.

Methods From our dedicated aneurysmal databank between October 1985 and July 2008, we reviewed 16 consecutive adolescents who had 20 intracranial aneurysms.

Results Ten boys and six girls (male/female ratio=1.67:1; mean age 16.78 ± 1.18 years) were included in the present study. Intracranial aneurysms in adolescents constituted 0.91% of all intracranial aneurysms. It was found that 25%

J.-t. Liang · Z.-y. Wang (⊠) Department of Neurosurgery, Peking University Third Hospital, No. 49 Huayuan North Road, Haidian District, Beijing 100191, China e-mail: wangzhenyu0001@yahoo.cn

J.-t. Liang · Y.-h. Bao · H.-q. Zhang · F. Ling (⊠)
Department of Neurosurgery, Xuanwu Hospital of Capital
Medical University,
45 Changchun Street,
Beijing 100053, China
e-mail: sdykdxxwyyhhll@yahoo.cn

J.-t. Liang · Y.-h. Bao · H.-q. Zhang · F. Ling Cerebrovascular Diseases Research Institute, 45 Changchun Street, Beijing 100053, China

L.-r. Huo

Key Laboratory for Neurodegenerative Disorders of the Ministry of Education, Capital Medical University, Beijing 100069, China of the lesions were in the posterior circulation, while 75% of the lesions were in the anterior circulation, and 25% developed on the middle cerebral artery (MCA). Half of the patients presented with subarachnoid hemorrhage and others mainly presented with mass effect such as weakness in the extremities, diplopia, and dysfunction of eye movement. Eight cases underwent endovascular treatment: including GDC therapy in five patients, parental artery occlusion in two patients, and cover stent implantation in one patient with pseudoaneurysm of the cavernous segment of the left internal carotid artery. Four patients received microsurgical therapy: aneurismal neck clipping for two patients and extracranialintracranial (EC-IC) bypass and trapping of complex aneurysms in MCA for the other two patients. Four patients did not receive microsurgical or endovascular therapy, including a boy whose aneurysm spontaneously thrombosed preoperatively and a girl who died before operation because of rerupture of aneurysm. Two patients did not undergo therapy owing to the high operative risk. All of the patients who received therapy had favorable outcome (GOS 4 or 5) at discharge and at follow-up.

Conclusions Intracranial aneurysms in adolescents differ from those in adults in many ways including the following: male predominance; high incidence of large or giant, traumatic, dissecting, and fusiform aneurysms; high incidence of aneurysms in the posterior circulation; high incidence of spontaneous thrombosis; better Hunt–Hess grade at presentation; and better therapeutic outcome. Both microsurgical approaches and endovascular treatment were effective. For some giant, complex intracranial aneurysms, parent artery occlusion or EC–IC bypass is the best treatment choice.

Keywords Adolescent · Aneurysm · Intracranial · Microsurgery

Introduction

Intracranial aneurysms are extremely uncommon in the pediatric population (≤ 18 years old), and their incidence increases with age [11, 28]. There are many differences in the epidemiology, clinical characteristics, morphology, size, location, and outcome in this group of populations compared with that in adults [2]. However, what are the clinical and radiological characteristics and surgical outcome of intracranial aneurysm in adolescents ($15\leq age\leq 18$)? In this study, we have tried to analyze data from adolescent patients in order to clarify the above characteristics.

Methods

Sixteen consecutive adolescents ($15 \le age \le 18$ years old; ten boys, six girls; male/female ratio=1.67:1; mean age 16.78± 1.18 years) who were treated for intracranial aneurysms in our institute between October 1985 and July 2008 were included in the present 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.

On admission and preoperation, all patients were graded according to the Hunt-Hess classification. Grade 0–3 was considered as good status and grade 4–5 was considered as poor status. Patients without a history of subarachnoid hemorrhage (SAH) were included in grade 0. Besides the Hunt-Hess classification, all of 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 their discharge to check whether there were residual aneurysms.

Thirteen patients were followed up postoperatively at 3, 6, and 12 months, and subsequently at yearly intervals; DSA was performed in 13 patients at 3 or 6 months to check if aneurysms had recurred. The mean follow-up was 19.2 ± 7.2 months (range 3–37 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 1–3 were considered as unfavorable.

Results

Neurological state on admission

According to the Hunt–Hess classification, eight were in grade 0, three were in grade 1, three were in grade 2, and two were in grade 3.

According to the Fisher grading system, eight patients were in grade 1 (no history of SAH), seven were in grade 2, and one patient was in grade 4.

Clinical presentation

The predominant clinical presentation of patients in our series included the following: sudden severe headache in eight patients due to SAH, vertigo in two patients, and diplopia and defect of eye movement in two patients. There was slight hemiparesis owing to cerebral infarction in one case and slight aphasia and hemorrhinia in another case, respectively. Two patients had no relative symptoms; they were detected because of head trauma (patient no.1) and pituitary adenoma (patient no.13).

Radiological characteristics

All patients underwent DSA to clarify the location, size, shape, and number of aneurysms. In our series, there were a total of 20 aneurysms in 16 patients (Table 1). On angiography, nine aneurysms were of the saccular type, seven were of the fusiform or irregular type, three were suspicious of infectious aneurysm and one was of pseudoaneurysm. There were 12 small, four large, and four giant aneurysms (small, diameter (Φ)< 10 mm; large, 10 mm $< \Phi < 25$ mm; giant, $\Phi > 25$ mm). Five aneurysms were located in the posterior circulation and 15 were in the anterior circulation. The middle cerebral artery (MCA) was the most common site of aneurysm (n=5): two aneurysms were located in the MCA bifurcation and three were in the distal MCA territory. In our case series, no aneurysms were located in the anterior communicating artery or anterior cerebral artery. There were three patients who had multiple aneurysms: two patients had two aneurysms and another patient had three aneurysms. The sites of aneurysms that were located in the posterior circulation included the basilar artery (n=3), vertebral artery (n=1), and posterior cerebral artery (n=1).

Therapeutic strategies

Eight patients underwent endovascular therapy, including GDC coiling in five patients, parent artery occlusion in two patients with giant aneurysms in the cavernous segment of the internal carotid artery (ICA), and placing cover stent for one patient with pseudoaneurysm. Four patients were treated by microsurgical technique: two by aneurysmal neck clipping and the other two by extraintracranial artery bypass.

Table 1 Clinical characteristics of 16 adolescents with 20 intracranial aneurysms

Case number	Sex	Age (years)	Accompanying disease	Initial symptom	Fisher	HHS	Site	Size	Therapy	Glasgow Outcome Scale
Patients	who u	nderwent	surgery							
1	М	15	Head trauma	None	1	0^{a}	R-PCom	11 mm	GDC coiling	5
2	F	15	None	Vertigo	1	0^{a}	BA bifurcation	6.3 mm	GDC coiling	5
3	F	15	CHD	Sudden headache	2	3 ^a	L-MCA	2 mm	GDC coiling	4
4	М	16	None	Diplopia	1	0 ^a	R-ICA-C4	35 mm	Parent artery occlusion	4
5	М	16	None	Sudden headache	2	1 ^a	R-MCA	20 mm	ECA–GSV–MCA bypass	4
6	Μ	17	None	Sudden headache	2	1 ^a	L-ICA bifurcation	12 mm	GDC coiling	4
7	М	17	None	Sudden headache	2	2 ^a	R-AchA	5 mm	Clip	5
8	F	17	None	Slight aphasia	1	0^{a}	L-MCA	42 mm	STA-MCA bypass	4
9	М	18	None	Diplopia	1	0^{a}	L-ICA-C4	60 mm	Parent artery occlusion	5
10	М	18	Head trauma	Hemorrhinia	1	0^{a}	L-ICA-C4 pseudoaneurysm	3 mm	Cover stent	5
11	Μ	18	None	Sudden headache	2	3 ^a	L-PCom L-AchA	6 mm 5 mm	Clip	4
12	F	18	None	Sudden headache	2	2 ^a	BA	8 mm	GDC coiling	5
Patients	who d	id not un	dergo surgery							
13	М	16	Pituitary adenoma	None	1	0^{b}	R-ICA-C2	7 mm		5
14	F	16	CHD, epilepsy	Cerebral infarction	1	0 ^b	L-MCA R-MCA R-PCA	3 aneurysmal enlargement		4
15	М	18	None	Vertigo	4	1^{b}	R-VA	41 mm		5
16	F	18	Left occipital AVM	Sudden headache	2	2 ^b	L-OphA BA	14 mm 7 mm	Pre-op death	1

L left, R right, HHS Hunt-Hess Scale, GOS Glasgow Outcome Scale, ICA internal carotid artery, MCA middle cerebral artery, PCoA posterior cerebral artery, BA basilar artery, AchA anterior choroidal artery, CHD congenital heart disease, GSV great saphenous vein, AVM arteriovenous malformation, OphA ophthalmic artery, VA vertebral artery, ICA-C4 cavernous segment of ICA, ICA-C2 superior clinoid segment of ICA

^a Hunt-Hess Scale before operation

^b Hunt–Hess Scale on admission

Four patients (with a total of seven aneurysms) did not receive surgical therapy:

The patient (no. 13) with a R-ICA paraclinoid aneurysm accompanied with pituitary adenoma had no relative symptom. DSA showed the fusiform aneurysm of irregular shape. His parents preferred further follow-up and refused surgery because of the operative risk invoved.

The patient (no. 14) had a history of congenital heart disease, cerebral infarction, and epilepsy; she had three small aneurysms (suspected as infectious aneurysm) located in the distal branches of the bilateral MCA and the right PCA and which were difficult to treat by endovascular or microsurgical method.

The patient (no. 15; Fig. 1), who had DSA in another hospital, demonstrated a giant dissecting aneurysm in the left vertebral artery. At 2 months later, a rechecked angiography in our institute showed that the parent artery and aneurysm regressed spontaneously prior to treatment. The patient (no. 16), who was with a history of subarachnoid hemorrhage for 10 days and whose Hunt– Hess grade on admission was 2, had both aneurysms in paraclinoid ICA and the original segment of basilar artery, respectively, associated with left occipital arteriovenous malformation (AVM). The aneurysm of basilar artery ruptured again preoperatively and the patient was Fig. 1 Patient 15: male, 18 years old. a Anteroposterior and b lateral view, respectively, showed a giant dissecting aneurysm in right vertebral artery; c anteroposterior and d lateral view, respectively, showed the parent artery and aneurysm spontaneous thrombosis prior to treatment



in a deep coma state and respiratory arrest at once. The patient died 2 days later.

Discussion

Intracranial aneurysms are much less frequent in children than in adult. The prevalence of pediatric aneurysms among all intracranial aneurysms ranged from 0.5% to 4.6% in previous reports [6, 17, 20]. A similar experience has been noted in a study by Kapoor [9] who, while examining 1,000 human cadaveric brains, found only one patient under the age of 20 years as harboring an intracranial aneurysm.

During the period of study, 1,750 patients, of all ages, with intracranial aneurysms were treated in our institute. Of these, 24 pediatric (age \leq 14) [15] and adolescent patients (15 \leq age \leq 18) were included, constituting 1.37% (24/1,750) and 0.91% (16/1,750) of the total number, respectively. So, the total incidence in patients under 18 years was 2.3% and thus similar to the above reports.

In children, male preponderance is seen [10], unlike the female preponderance in adults [1]. This male dominance was also found in our study. The ratio between males and females was 1.67:1 (M/F=10:6), which is similar to what was reported by Huang [8].

The presenting features in children differ from those in adults. The incidence of SAH in various series varied from 35% to 100% [8, 23, 25]. In this series, half (50%) of the patients presented as SAH and the other half of patients had presentations other than SAH, including proptosis, diplopia, hemiparesis, chronic or intermittent headache, and vertigo and so on. In our series, all of the cases had good Hunt-Hess grades (I-III) at presentation, whereas only 42% cases had a good grade in the series by Huang [8]. The higher incidence of good grade in our series may be due to the fact that the patients with poor grade never reached the hospital. Aneurysms in the pediatric age group tend to be large in size. The incidence of giant aneurysms in children varies from 3% to 37% [7, 25]. In the present series, 20% of the aneurysms were giant (Φ >25 mm), and 20% of the aneurysms were large (10 mm $< \Phi < 25$ mm). The large size of aneurysms is partly responsible for the large number of patients presenting with mass lesions. The incidence of multiple aneurysms in various series is 4-15% [6]. In the present series, 18.8% (3/16) of adolescents had multiple aneurysms.

The location of the aneurysms is also different in children. According to most literature, the most common site for pediatric intracranial aneurysms is the ICA bifurcation, with the incidence ranging from 24% to 50% [16, 21, 22]. However, we did not find this predilection for ICA bifurcation site in our case series. Only one (5%) aneurysm occurred at the ICA bifurcation. This was inconsistent with the above reports. The MCA was the most common site for aneurysms (n=5). Two aneurysms were located in the MCA bifurcation and three were in the distal MCA territory. Three were also in the cavernous segment of the ICA and another three were in the basilar artery. According to the literature, the higher incidence of posterior circulation aneurysms is also a characteristic of the pediatric population [8]. In our study, 25% (5/20) of the aneurysms were located in the posterior circulation,

including the basilar artery (n=3), vertebral artery (n=1), and posterior cerebral artery (n=1).

The high incidence of traumatic aneurysms is another characteristic in the pediatric patients. Traumatic aneurysms account for 14–39% of all the pediatric aneurysms [13, 29]. In the present study, there was one traumatic and pseudoaneurysm. Nakstad [19] described the typical features of these traumatic aneurysms—tending to be supratentorial, peripheral, irregular, lacking a proper neck, and arising at sites other than arterial bifurcations. He also reported 60 traumatic aneurysms, and 75% of them were in children. So, if children with closed head injury showed unexplained neurological deterioration, failed to improve as expected, or developed new neurological symptoms or signs, they should undergo diagnostic imaging.

In our case series, there was one patient whose aneurysms and parent artery had complete spontaneous thrombosis before treatment. The incidence was 1/16 (8.3%). In a report by Lasjaunias [12], ten patients had complete or partial spontaneous thrombosis among a total of 59 pediatric intracranial aneurysm patients. Unfortunately, the exact



Fig. 2 Patient 8: female, 17 years old. A giant aneurysm in the left MCA was treated by STA-MCA bypass. **a** Anteroposterior and **b** lateral view, respectively, showed the aneurysm in the left MCA preoperation; **c** anteroposterior and **d** lateral view, respectively, showed

the MCA and aneurysm dismissed post-operation; \mathbf{e} lateral and \mathbf{f} anteroposterior view, respectively, showed that the MCA territory was supplied by ipsilateral ECA

mechanisms and factors responsible for this occurrence cannot be predicted with sufficient reliability. Liang et al. [15] have also reported the same outcome: two patients had complete spontaneous thrombosis among a total of 24 pediatric patients.

With regard to treating aneurysms, the goal should be not only to achieve alleviation of the acute symptoms but also, most importantly, to protect the patient from future bleeding. For adolescents with intracranial aneurysm, the factors involved in treatment selection are considerably different from that for adult patients. Concerns about safety and efficacy are uniform in both age groups, but the durability of treatment is also an important factor for adolescents for whom the remaining life expectancy is five to seven decades [25]. Sanai considered that microsurgical therapy led to a higher rate of complete obliteration and a lower rate of recurrence of aneurysm. So, he preferred this therapy for adolescent patients [25]. Proust also insisted the same treatment selection: among 22 children in their series, a microsurgical procedure was performed in 17 (77.3%). The endovascular approach was used only in four patients (18.2%) and a combined approach was used in one (4.5%)[23]. However, with the development of endovascular technology and device in the last decade, endovascular therapy had improved the safety, efficacy, and durability for aneurysm. After all, the endovascular approach was more microinvasive for adolescents.

The treatment selection for a given aneurysm in our institution is made by a multidisciplinary team of neurosurgeons and interventional neuroradiologists. In our study, the vast majority (eight out of 12) of patients who underwent surgical therapy received endovascular treatment and all had good recovery. Only four patients underwent microsurgical therapy.

Extracranial–intracranial (EC–IC) bypass is an important selection in the treatment of patients who require therapeutic occlusion of large intracranial vessels, such as the ICA or proximal MCA, for the management of complex intracranial aneurysms [3, 4, 14, 27]. Techniques such as in situ anastomosis, EC–IC arterial grafting, and great saphenous vein or radial artery grafting have been applied to both the anterior and posterior circulations [5, 18]. The reported overall complication rate for EC–IC bypasses was low, with 10-year patency rates as high as 73% [24].

In the present study, both of the cases (case 5 and case 8) with complex MCA aneurysms were treated by EC–IC bypass. Patient 5 was a 15-year-old boy who presented with sudden headache. Computed tomography (CT) showed subarachnoid hemorrhage and cerebral angiography demonstrated a 18×20-mm complex right MCA aneurysm. Patient 8 was a 17-year-old girl who initially presented with slight right hemiparesis and aphasia (Fig. 2). CT showed a huge mass in the left Sylvian fissure and cerebral

angiography demonstrated a 42×36 -mm giant, complex left MCA aneurysm. The distal branches of MCA were all originated from the aneurysm in both cases and balloon occlusion test showed that the collateral circulation was not sufficient. So, we trapped the aneurysm and affected MCA segment, combined with an autogenous saphenous vein graft bypass from the cervical external carotid artery to the distal MCA, for patient 5. For patient 8, she underwent EC–IC bypass from the superficial temporal artery (both frontal and parietal branches) to both of the distal branches of MCA.

In our case series, for the management of complex intracranial aneurysms, the indications for combining cerebral revascularization with proximal ligation or trapping of the offending vessel segment were on the basis of the patient's collateral circulation and tolerance to afferent vessel occlusion, which is similar to those described by [4, 26].

Conclusions

Intracranial aneurysms in adolescents have many clinical and radiological characteristics that differ from the intracranial aneurysms in adults: male predominance; high incidence of giant, traumatic, dissecting, and fusiform aneurysms; high incidence of aneurysms in the posterior circulation; high incidence of spontaneous thrombosis; and better Hunt–Hess grades at presentation. Intracranial aneurysms in adolescents also have a better therapeutic outcome. For intracranial aneurysms in adolescents, both the microsurgical approaches and the endovascular treatment were effective. For giant, complex intracranial aneurysms, parent artery occlusion or EC–IC bypass is the best treatment choice.

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