

Aneurysms in childhood: literature review and cases description

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Introduction: In the aneurysm disease, children aneurysms do not represent a majority of the diagnosis. Associated conditions should be monitored, including aortic coarctation, polycystic kidney disease, fibromuscular dysplasia, tuberous sclerosis, Ehlers-Danlos syndrome, and Marfan syndrome. They can present as congenital, infectious, or traumatic.

Case report: We describe a series of five cases of aneurysms in patients ranging from 15 to 17 years of age whose aneurysms were found on examination. In most cases, patients were asymptomatic and did not have predisposing syndromes. Surgery was the treatment of choice in all cases. No patient had a recurrence in the following years of radiographic follow-up.

Methodology: This is a literature review and case series analysis with a narrative-descriptive approach. The database for the search was PubMed®, which were used to search for articles with the subject descriptors brain, aneurysms, pediatric and child. Between the descriptors, the boolean operator and was used.

Results: Aneurysms can be classified by their size and morphology, as well as having a predilection for anterior circulation. The most common symptom observed in our pediatric series of 5 patients was migraine or chronic headache (60%), followed by asymptomatic patients (40%). Three of five patients (60%) in this study were previously hypertensive without other comorbidity, while 40% of the cases were totally healthy.

Discussion: Pediatric intracranial aneurysms are relatively rare diseases. They require accurate and detailed diagnosis, which need to be assessed and managed in a multidisciplinary team.

Keywords: aneurysm, cerebral, child

INTRODUCTION

Cerebral aneurysms in children are not the majority diagnosis of this disease, representing 1.6 to 7% of all intracranial aneurysms [1,3,12,16,17]. It is estimated that there are approximately 1 to 3 cases of a childhood aneurysm per 1 million population [1]. One of the first documented cases in children was a 15-year-old boy, as reported by the German pathologist Eppinger in 1871 [4].

The diagnosis was made on the basis of an autopsy, which revealed a constriction of the aortic isthmus and an unusual finding of free blood at the base of the brain, blood resulting from the rupture of a saccular aneurysm of the right anterior cerebral artery. The first alive case was reported by Edvard Bull of a 17-year-old girl who presented a severe headache and oculomotor nerve paralysis, being diagnosed with a ruptured posterior communicating artery aneurysm, confirmed by the autopsy posteriorly [1,4]. Ruptured infantile aneurysms account for a mortality rate between 10-23% [2]. An important, but not only, causal conditional of



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aneurysms are endothelial injury and transmural vascular dissection that weakens the adventitial and muscular layer, and because it is a rare pediatric condition, associated conditions must be monitored, including aortic coarctation, polycystic kidney disease, fibromuscular dysplasia, tuberous sclerosis, Ehlers-Danlos syndrome, Marfan syndrome, immunodeficiencies, and autoimmune diseases [1,2].

Although in the adult population several risk factors may contribute to aneurysm formation, they are unlikely to contribute to aneurysm formation in the pediatric population [3]. A number of authors have postulated a greater role for congenital factors [1,2,3] in the development of childhood aneurysms, in addition to traumatic and infectious factors [1,4]. Their clinical presentation includes blurred or double vision, confusion, nausea and vomiting, seizures, severe headache, loss of consciousness, droopy eyelid, and stiff neck.

Aneurysm should be a differential diagnosis in patients with this presentation, and when suspected, the patient should be promptly evaluated by a physician and undergo computed tomography (CT) or magnetic resonance angiography (MRA), and a gold standard four-vessel cerebral angiography (DSA).

The anatomical and topographical characteristics of childhood aneurysms differ from those of adults, have a male predilection, mostly single presentation, and become symptomatic from birth to 6 years of age and from 8 years to adolescence, but their clinical manifestations and complications, such as aneurysmal subarachnoid hemorrhage (aSAH) are relatively poorly understood.

We present a case series of 5 patients who attended our services and were diagnosed with aneurysm and treated accordingly, as well as review the most recent literature.

CASE REPORT

Case 1

Male, 16 years old, with symptoms of chronic headache. During the investigation, an anterior communicating artery aneurysm was found, measuring about 3mm (show in fig. 1). The patient had no predisposing personal history, was previously healthy, without comorbidities, without Ehler-Danlos, Marfan and polycystic kidney. As surgical methods, we used a pterional craniotomy with high surgery, without postoperative and/or disclosed signs. There was no recurrence of aneurysms in a 10-year follow-up.

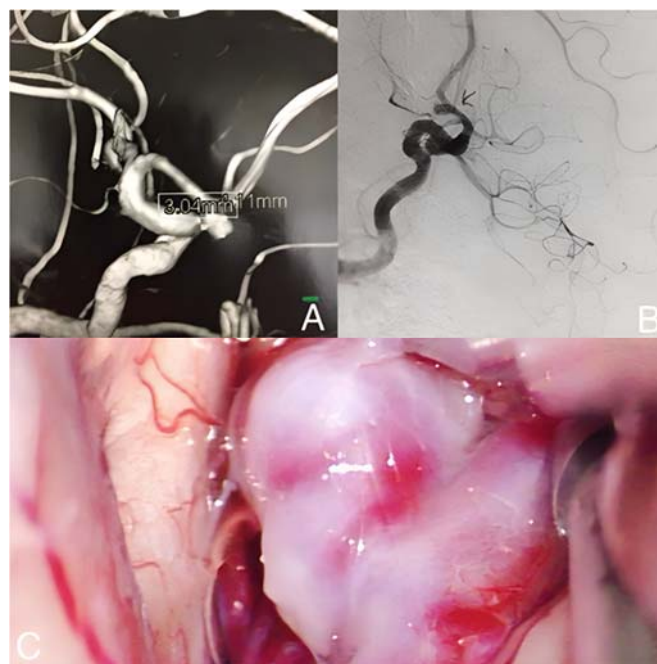


Figure 1 - A- Anterior communicating artery aneurysm with bottom pointing forward and downward in 3D reconstruction angiography. B- Anterior communicating artery aneurysm in Angiography. C- Intraoperative anterior communicating artery aneurysm.

Case 2

Female, 15 years old, reporting only a migraine complaint. During the investigation, a posterior communicating artery aneurysm was identified. Patient presented digital clubbing, indicating a marfanoid profile. In the immediate postoperative period, the patient has no neurological deficits. Patient remains unchanged 8 years after surgery (show in fig. 2).

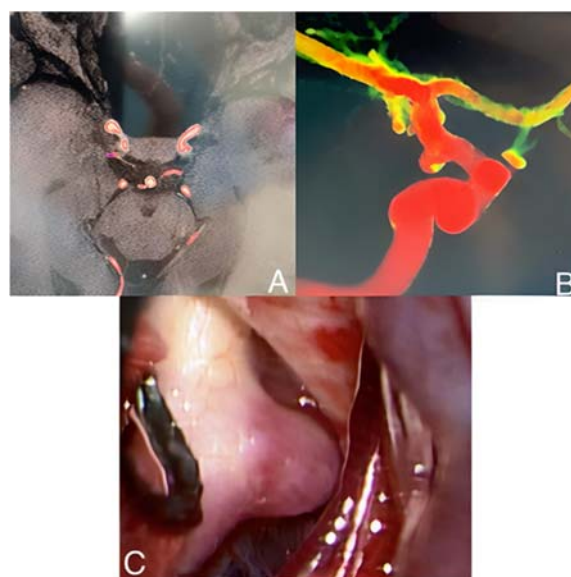


Figure 2 - A- A posterior communicating artery aneurysm found in arterial angiotomography. B- A right posterior communicating artery aneurysm found in arterial angiotomography. C- Intraoperative posterior communicating artery aneurysm.

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Case 3

Female, 17 years old, previously hypertensive. Examination findings revealed aneurysm in the choroidal artery and in the right middle cerebral artery (show in fig. 3). The surgery was performed with evoked potential and indocyanine, without major bleeding and complications. Postoperatively without deficits, the patient remained without deficits during the 7 years of follow-up.



Figure 3- A- Aneurysm in the right anterior choroidal artery and in the right middle cerebral artery in angiography. B- Aneurysm in the right anterior choroidal artery in 3D reconstruction angiography.

Case 4

Male patient, 16 years old, precisely hypertensive and using chemotherapy to treat chronic myeloid leukemia. In an investigation of chronic headache, a giant aneurysm was found in the carotid bifurcation (show in fig. 4). Clipping of the right aneurysm was uneventful. The patient evolved with left hemiparesis in the postoperative period, fully recovering after 6 months of the surgical procedure. Patient remains without deficits after 5 years of follow-up.

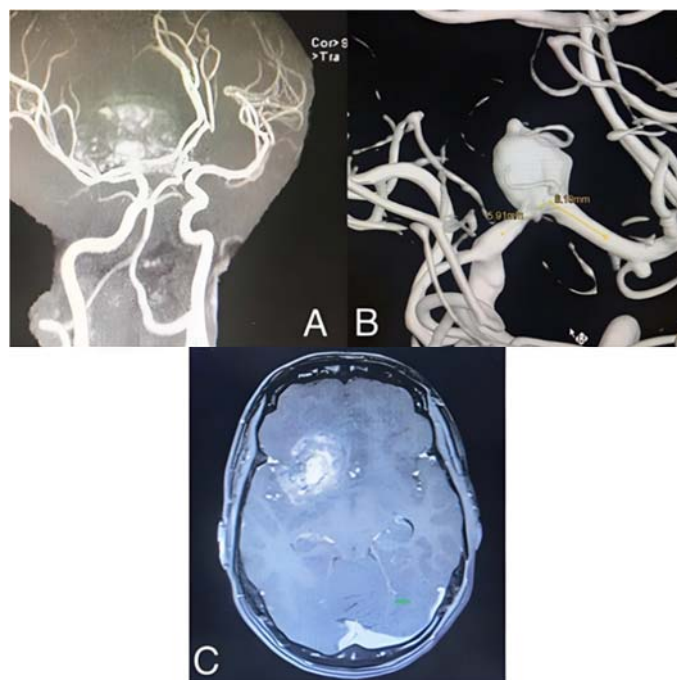


Figure 4- A- Giant aneurysm located in the internal carotid bifurcation with compression in lower portion of frontal lobe near the Sylvian fissure. B - Giant aneurysm in the carotid bifurcation found in angiography. C- Giant aneurysm was found in the right carotid bifurcation in angiography.

Case 5

Female patient, 17 years old, previously hypertensive. Two aneurysms of the choroid artery and of the right middle cerebral artery were found on examination (show in fig.5). Surgery performed with evoked potential, without intraoperative bleeding. The patient evolved well, without major complications in the immediate postoperative period.



Figure 5- Aneurysms of the right choroid artery and of the right middle cerebral artery in 3D reconstructed angiography.

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METHODS

This is a literature review and case series analysis with a narrative-descriptive approach. The database for research was PubMed®, which used the subject descriptors cerebral, aneurysms, pediatric and child to search for articles. Between the descriptors, the boolean operator and was used.

The time interval was 5 years. The search was performed in January 2022, in free full text quality.

Only reviews and systematic reviews that covered the selected subject descriptors, were written in English, and whose journals were classified as qualis A or B according to CAPES were selected. Qualis C or unrated articles were excluded.

Finally, the articles were read to verify their consistency with the theme developed for the literature review.

The search in the data platform resulted in a total of 31 articles, of which 29 were classified as reviews and 2 as systematic reviews. After reading the articles to evaluate the link between descriptors, excluding the casual encounter of the searched terms, there were 17 reviews and 1 systematic review, with a total value of 18 articles used [Table 1].

Table 1- General data of the articles included in this review

	Reviews	Systematic Reviews	Overall
Found articles	29	2	31
Selected articles	17	1	18

DISCUSSION

Aneurysms can be classified by their size, being small (< 5mm), large (6-24mm) or giant (> 25mm), morphology, divided into saccular, fusiform or dissecting [1,2,3], and also if they're presented single or multiple (only in 10% of the cases 2). Table 2 presents specific data of recent largest series of pediatric aneurysms' cases.

Aneurysms are preferably located in the anterior circulation (75% of cases), with the internal carotid artery, middle cerebral artery and anterior cerebral artery being the most frequent locations, and posterior circulation in 25%, more often in the basilar artery [2,3]. Location and size are not similar in pediatric and adult patients. A recent study conducted by Chen, R. et al (2021) showed that 24.5% of pediatric aneurysms are giant; this data corroborates robust other literatures that links 20 to 45% of childhood aneurysms as giant, unlike adult cases that have the giant ones as representative in less than 5%.

If the intracranial aneurysms ruptures, fatal conditions in clinical settings such as subarachnoid hemorrhage (SAH) or intracerebral hemorrhage (ICH) could occur, with 1-year mortality rates up to 65% [11,12].

According to Chen, R. et al (2021), cited before, the most common symptom observed in the pediatric series of 94 patients was headache (58,5%), followed by vomiting (48,9%), seizure (34%) and loss of consciousness (14,9%). In comparison to our series, 60% of the cases had chronic headache or migraine, while 40% were asymptomatic and the aneurysm was an examination finding.

Our number of cases may not be as expressive as other comparative studies in order that these patient samples correspond to an adult hospital service, not specialized or referenced in child/pediatric treatment. Also, comparing to the most representative recent series, our patients are in second childhood (15 to 17 years old), however, Matson's landmark series of pediatric aneurysms [19] contained no patients younger than 16 years of age, which did not underestimate the value of his literature.

Pediatric and adult intracranial aneurysms differ from several characteristics, such as lifestyle risks (smoking, alcohol consumption) and previous health conditions (hypertension, diabetes mellitus, migraine and hypercholesterolemia). Children are far less exposed to lifestyle risks as adults [14] and, moreover, those with previous health conditions are significantly lower than in adults [15].

Though, among pediatric cases, the risk factors for rupture are divided in different groups: age below 5 years old and aneurysms located in the distal arterial region (DAR) increases the risk of rupture, while wide neck aneurysms could be a protective factor in the pediatric age group, all factors independently [13].

They require accurate and detailed diagnosis, as they may arise from a variety of different underlying pathological mechanisms, which need to be assessed and managed in a multidisciplinary team [1,2,4] to institute the correct treatment strategy.

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Table 2- Largest recent series of pediatric aneurysms

	Hetts et al, 2009 [20]	Koroknay-Pál et al, 2012 [21]	Mehrotra et al, 2012 [22]	Chen et al, 2021 [23]
PATIENTS (N)	77	114	57	94
NUMBER OF ANEURYSMS	103	130	73	-
AGE (RANGE)	12 y (3 mo-18y)	14.5 y (3 mo-18y)	12.7 y (4y-18y)	10.6 y (21d-17y10m)
SEX	F/M 1.1:1	M/F 3:2	M/F 1:1.2	M/F 2.4:1
MORPHOLOGY				
FUSIFORM (%)	31	10	2.7	-
SACCULAR (%)	46	78	78	-
INFECTIOUS (%)	12	0	2.7	-
TRAUMATIC (%)	14	7.6	8	-
GIANT (>25MM) (%)	11	12	19	24.5
MULTIPLE (%)	16	11	11	2.1
ANTERIOR CIRCULATION (%)	78	89	71.3	87.2
POSTERIOR CIRCULATION (%)	22	11	28.7	12.8
SAH (%)	32	78.1	88.7	60.6
MASS EFFECT (%)	-	6.1	7	-
MORTALITY (%)	1.3	7.7	8.7	-

Treatment options for aneurysm include endovascular or surgical approach. Each has its own advantages and disadvantages, but there's no available literature that demonstrates superiority on either one of the approaches on long term clinical outcomes when children are treated, including ruptured or unruptured aneurysms. Though, the assistant physician may access either modality based on its expert clinical judgment, considering patient preference, local expertise and aneurysm characteristics [18]. In our service, the preferred modality is aneurysm clipping as in the cases presented before. New prospective studies are

valuable to establish benefits of either definitive treatment for aneurysms in the pediatric population.

CONCLUSION

Pediatric intracranial aneurysms are relatively rare diseases. They require accurate and detailed diagnosis and the correct approach is to be managed by a multidisciplinary team. Available literature shows that there was no statistically demonstrable difference between endovascular and surgical treatment. The present article consists of a case

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series (corresponding to an adult hospital service), taking into consideration reviews already published; however, even the available literature lacks information when compared to adults.

Ethical approval

This study was performed in line with the principles of the Declaration of Helsinki. Approval was granted by the local Ethics Committee

Consent to participate

The patient gave consent to use his information and images for research proposes.

Consent for publication

The patient gave consent to use his information and images for publication.

Conflict of interest

The authors declare no conflicts of interest with respect to the content, authorship, and/or publication of this article.

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