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Research Article

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Endo Vascular Management of Supra and Infra Tentorial Pediatric Intracranial Aneurysms; A Single Center Experience

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Abstract

Intracranial aneurysms are rare in pediatric population. They have different pathophysiology, location, type, outcome than adults. Long life expectancy, denovo occurrence and their tendency to recur necessitate the long term follow up in these cases. Here we summarize our experience in treating these aneurysms in a tertiary care hospital from 2014 to 2021. We found 4% of our total aneurysm population to be under 18 years with a male to female ratio of 1.5:1. 13.3% patients had multiple aneurysms and 38.9% had posterior circulation aneurysms. Most common site of aneurysm was ICA bifurcation. Most of the patients were treated endovascularly with favorable outcome at follow up.

1. Introduction

Intracranial aneurysms (IA) are uncommon in pediatric population as they represent less than 5% of all intracranial aneurysms [1]. But unlike adults, they differ in their etiology, pathogenesis, aneurysm types, location, presentation and outcome [2]. Aneurysms in posterior circulation and terminal internal carotid artery (ICA) are more common in pediatric groups than in adults [1,2]. Dissecting and fusiform aneurysms are more commonly encountered than in adults [1]. Furthermore, aneurysms tend to be larger with more tendencies to have spontaneous thrombosis than adults [1]. The children present with unusual headache, subarachnoid hemorrhage (SAH), cranial nerve palsy, symptoms related to raised intracranial pressure or a combination of these [3]. High index of suspicion can lead to prompt diagnosis through cranial and vascular imaging by CT or MR. Considering the longer life expectancy, IA in pediatric groups have higher recurrence rate and more de novo formations [4]. So long term follow-up schedules should be ensured.

2. Materials and Methods

Children of less than 18 years of age who were diagnosed and managed for intracranial aneurysms over a period of 2014 to 2021 were included in this study. The data were retrospectively reviewed from the database of our hospital. Demographics, clinical information, radiological findings, treatment and outcome were analyzed. Patients who were diagnosed to have intracranial aneurysms on the basis of four vessel intra-arterial digital subtraction angiogram and underwent clipping or endovascular coiling or conservative management were included in the study. Hunt and Hess Grade was used to grade the subarachnoid hemorrhage. A grade 0 was assigned to patients with unruptured aneurysms. A grade I to III was considered as a good grade SAH, whereas grade IV –V was considered as poor grade SAH. CT scan followed by DSA was done in all patients. The aneurysms were subdivided on basis of size into the following pattern – small(<10mm), large(10-25mm) and giant (>25mm).Once the aneurysm was secured by either coiling or clipping the management of SAH was consistent with the standard guidelines. Patients were followed up after a period of 3 to 6 months after discharge from hospital.Post coiling DSA was done at 3 months. Glasgow outcome score (GOS) was used to assess post-coiling or post-operative outcome and follow up.

3. Results

At our center, out of 367 patients with intracranial aneurysms treated between 2014 and 2021, 16 patients were less than 18 years which accounts for about 4.4% of total population. The age group ranged from 4 years to 18 years with mean age of about 14.8 years and median age was 15.5 years. 1 patient was below 5 years. Male to female ratio of 1.67:1 (table 1)

Age group (years)	Number of patients
5 or less	1
6 to 14	7
15 to 18	8

Table 01: Age distribution

4. Presentation

Headache was the most common presenting feature of around 80% patients. Five patients (31.3%) had ictal loss of consciousness, 5(31.3%) had seizure and 2 (12.5%) had hemi paresis. Cranial nerve involvement was seen in 3 patients (18.8%) with the 3rd nerve palsy being the most commonly. Other presenting

symptoms include fever, vomiting (table 2).

Symptoms	Number of patients
Headache	13
Seizure	5
Loss of consciousness	5
Hemi paresis	2
Cranial nerve involvement	3
Fever	2

Table 02: Presenting symptoms

Hunt and Hess grading of patients are given in Table 3.Grade 1 was the most common grade seen in 6 patients. Good grade SAH seen in 13 (86%) patients.

Hunt and Hess grade	Patients (%)
0	3(18.8%)
1	6(37.5%)
2	3(18.8%)
3	2(12.5%)
4	1(6.3%)
5	1(6.3%)

Table 03: Hunt and Hess grade

CT finding	Number of Patients
SAH	11
IVH	01
ICH	01
No hemorrhage	03

Table 04: CT findings of patients

Traits	Description	Number of aneurysms
Aneurysm location	Anterior circulation	11
	Posterior circulation	08
Side of aneurysm	Right	05
	Left	10
	Bilateral	01
Site of Aneurysm	ICA –Cavernous segment	01
	ICA bifurcation	06
	MCA	03
	ACOM	01
	Superior cerebellar artery	01
	Basilar artery	03
	PICA	02
	PCA	01
	VA	01
Types of Aneurysm	Saccular	11
	Dissecting	06
	Mycotic	01
	Fusiform	01
Size of Aneurysm	Small	14
	Large	02
	Giant	03

Table 05: Description of aneurysm

Treatment Modality	Number of Aneurysms
Simple Coiling	08(42.1%)
Glue Embolization	02 (10.5%)
Stent assisted coiling	02 (10.5%)
Balloon assisted coiling	01 (5.3%)
Flow diverter coiling	01 (5.3%)

Table 06: Endovascular treatment of aneurysms

5. Radiological Findings

In our series CT scan revealed subarachnoid hemorrhage in 11 patients (68.8%). Intra cerebral hemorrhage (ICH) with intra ventricular extension seen in 2 patients (12.5%) and rest of the patients had no intracranial bleed. Digital Subtraction Angiogram(DSA) in 16 patients revealed 19 aneurysms. 11 (57.9%) were in anterior circulation aneurysm and 8(42.1%) were posterior circulation aneurysm. ICA bifurcation was the commonest site (36.8%) of aneurysms in anterior circulation. Middle cerebral artery aneurysm was noted in 3(15.8%) patients. Among the posterior circulation aneurysms left side was more commonly involved than right side (1.8:1). Multiple aneurysms were seen in 2(12.5%); 1 had bilateral ICA aneurysm and other had aneurysms in the posterior circulation.

According to size, in our series 14 (73.7%) were small, whereas large and giant aneurysms noted in 2 (10.5%)and 3(15.8%) patients respectively. Saccular aneurysm were seen in 11 (57.9%), dissecting aneurysm were seen in 6 (31.6%), 1 (5.3%) was fusiform in nature and 1 (5.3%) was mycotic. Predisposing factors like polycystic kidney disease and coarctation of the aorta was seen in 2 patients.

6. Treatment

Endovascular intervention was done in 13 (81.5%) patients, while surgical intervention was done in 1 (6.2%) patient. 2 patients did not receive any modalities of treatment offered due to their financial constraints. Among total of 19 aneurysms; 8 (42.1%) aneurysms were treated with simple coiling, among these 6 were saccular and 2 were dissecting type of aneurysms. Glue embolization was done in 2 (10.5%) cases. Among them 01 was mycotic in nature in distal MCA and the other was small dissecting aneurysm in PICA. Stent assisted coiling was done in 2 (10.5%), balloon assisted coiling done in 1 (5.3%) patient and flow-diverter assisted coiling done in 1(5.3%) patient with giant saccular ICA bifurcation aneurysm. Stent assisted coiling was done in case of ICA bifurcation saccular aneurysm with distal ICA-MCA M1stenosis and giant aneurysm in left vertebral artery. Balloon assisted coiling was done for small dissecting aneurysm in left vertebral artery-basilar artery junction.

7. Complications

Vasospasm was seen in 6 patients, which were confirmed by angiography and intra-arterial nimodipine was administered in patients with severe vasospasm. One patient developed arterial infarcts during the course of illness. Four patients developed hydrocephalus and among them 2 patients required lumbar drainage for 3 consecutive days. One patient developed meningitis and improved with broad spectrum antibiotics. Other infective complications (including chest infection, urinary tract infection) were seen in 3 patients. No patient died on follow up after receiving treatment for aneurysm.

8. Outcome

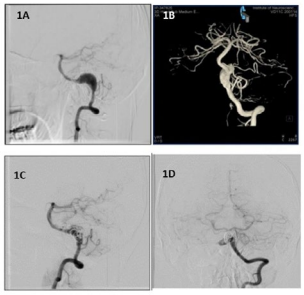
Among 14 patients who were treated at our institute; a favorable outcome was seen in 12 (87.5%) patients. Among them 9 (56.3%) patients were fully independent at the time of discharge. Three patients (18.8%) showed improvement in GCS but functionally independent after rehabilitation in the follow up. 2 (12.6%) patients were functionally dependent during the follow up.

9. Discussion

Intracranial aneurysms are uncommon in pediatric population. Multiple factors contribute to the development of pediatric IAs including trauma, infection, congenital vessel abnormalities, connective tissue disorder etc [19]. Hetts and colleagues grouped these factors into 'direct' primary triggers (like trauma, infection) or 'silent' genetic factors [21]. Lasjaunias and colleagues commented the pathogenesis of these IAs to be due to 'defective defense mechanism' and 'offensive etiological factors [22]. Vessel wall dysfunction, systemic or genetic disease lead to transient or permanent failure of repairing partial insults, termed defective defense mechanism [11]. Disorders associated with these IAs include: polycystic kidney disease, fibro muscular dysplasia, tuberous sclerosis, arteriovenous malformation, vascular anomalies, cardiac myxoma, aortic coarctation, cerebral tumors, Ehlers–Danlos syndrome, Marfan syndrome, irradiation, Moyamoya syndrome, human immunodeficiency virus, syphilis, thalassemia, glucose-6-phosphate dehydrogenase deficiency, sickle cell anemia, pseudoxanthoma elasticum etc [23]. Infectious IAs can be seen in distal small arteries when the septic emboli lodge in the endothelium of small arteries and infectious agents spread from vessel lumen to the extra-vascular space [25]. Larger arteries are affected when, infection extends from outside towards the lumen of the vessel or infectious emboli originating through vasa-vasorum [6]. Infectious agents can be found in blood or cerebrospinal fluid but in more than one-third cases they may not be detected [6]. Lashaunda's and colleagues categorized IAs into four types on the basis of angiographic findings and clinical scenario [11].

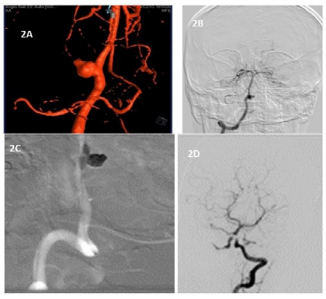
(1) Dissecting aneurysm without features of infection or trauma.
(2) Saccular aneurysm without features of infection or trauma.
(3) Infectious/Mycotic aneurysm in case of associated systemic infection or immunocompromise.
(4) Traumatic aneurysm with history of significant trauma [11].
According to size, IAs can be grouped into small (less than 10mm), large (10-25mm) and giant (more than 25mm) aneurysm [3]. Dissecting IAs are usually fusiform and giant aneurysms that undergo spontaneous thrombosis [11]. They account for about 16–45% of pediatric IAs1.Krings and colleagues reported that the incidence of dissecting aneurysms are four times higher in children than in adults [6]. We found 21% of our patients to have dissecting IAs. Spontaneous thrombosis of IAs occurs in about 8.3 to 16.9% of all cases1.Angiographically dissecting aneurysms can be identified by regular or irregular fusiform dilatation usually preceded by a stenosis that marks the proximal part of the dissection [6]. Saccular or 'Berry' aneurysms are rarely seen in children below 10 years of age, but can be found in up to 20% of cases below 18 years [3,11]. They are classically located at the vessel bifurcation6. ACOM, MCA bifurcation, basilar top is usual site of saccular aneurysm [12]. Infectious IAs account for about 15% of pediatric aneurysms6. The culprit organism here is mostly Staphylococcus aureus, followed by Streptococcus viridians and other gram-negative organisms [13]. These IAs

are usually located at the distal small arteries or near the skull-base6. Sometimes these can be multiple, especially in immuno-compromised patients [14]. On angiography they are often seen as small aneurysms arising from distal vessels extending into the hematoma [6,14]. Traumatic IAs is encountered in about 5-10% of all pediatric aneurysms [15]. 40% of these cases are found in distal ACA near falx cerebri, 35% cases in vessels of skull base and rest of the time in distal cortical vessels [6,15,16]. Usually 2-4 weeks after enduring a closed head injury, children present with intra cerebral hemorrhage [6].



Case 1: Figure 1A, 1B: Pre-operative angiogram revealed fusiform aneurysm involving v4 segment of vertebral artery
Figure 1C: Post-operative angiogram showed stent assisted coining of aneurysm
Figure 1D: Follow up angiogram

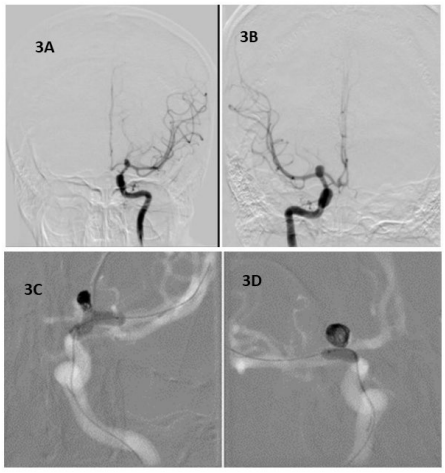
Intra cranial aneurysms (IAs) represents only 0.5 to 4.6% in pediatric population, with male to female ratio of 1.1:1 to 2.8:1 (1,5). In our series they constitute of about 4.4% of all aneurysms treated at our center. Our series has male predominance, with male to female ratio was 1.67:1, consistent with literature. The male predominance may be due to the fact that, around one third of pediatric IAs is caused by trauma and males are more prone to it [2]. Pediatric intra cranial aneurysms differ from adult counterpart by tendency to have larger aneurysm, higher incidence in posterior circulation and ICA bifurcation, complex structure and recurrence [2,6].



Case 2: Figure 2A, 2B: Pre-operative angiogram revealed saccular aneurysm of basilar artery
Figure 2C: balloon assisted coiling of the aneurysm
Figure 2D: Follow up angiogram after 3 months

10. Location

The site of aneurysm differs from that of adults. The most common location of pediatric IAs is terminal ICA [12]. But other common locations include ACA, MCA, ICA and basilar artery [11,12,17]. Posterior circulation aneurysms are three times more common in pediatric group than in adults [5,18]. The reported incidence of MCA IAs is 18% of all for both adults and children [18]. But most neonatal IAs are found in distal circulation, predominantly the MCA territory19. For giant IAs, incidence varies between 12% and 37% among different case series [20]. There are variations of IA among different age groups of pediatric population [6]. Dissecting IAs is commoner before 5 years of age, while classic saccular IAs occur later in life [6]. In our series, 11 (57.9%) were saccular aneurysm and 6 (31.8%) were dissecting aneurysms. Most of our patients (93.8%) age ranged from 12-18 years, which might explain the higher incidence of saccular IAs in our series. Middle cerebral artery (MCA) aneurysms and posterior communicating artery (PCOM) aneurysms are also less commonly found than adult [6]. But, multiple aneurysms are twice as more common in pediatric group than in adults [8]. In our series we found 12.5% of our patients having multiple IAs. The anterior communicating artery (ACOM) aneurysm is rare in infants but becomes the most common site after the age of 15 years7. Only one of our patients had ACOM aneurysm who was 16 years of age.



Case 3: Figure 3A, 3B: Pre-operative angiogram revealed saccular aneurysm involving bilateral internal carotid artery bifurcation
Figure 3C: balloon assisted coiling of both the aneurysms
Figure 3D: post-operative angiogram following coiling of both the aneurysms
Figure 3E, 3F: Follow up angiogram after 3 months

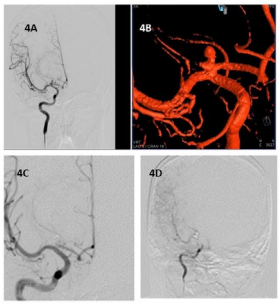
11. Presentation

Children with IAs usually present with headache, SAH, focal neurological deficits, compressive features, seizures, ischemic features or a combination of these [2,3,6]. But non-hemorrhagic presentation is more common than adults [9]. SAH as a presenting feature of pediatric IAs ranges from 58 to 91 % among different literatures [10]. In our series; 68.8% patients presented with SAH. Children presents with better clinical grades than adults with SAH [2]. It may be due prominent lepto-meningeal collaterals, fewer co-morbidities, increased activity of nitric oxide pathway etc [2,11]. Seizure and head ache as a presenting feature of IAs, is commoner in children than in adults [2,10].

But as these aneurysms become larger, compressive features like cranial nerve palsy, focal neurological deficits also become evident. Interestingly, cerebral infarction can also be a presenting feature of IAs, specially in case of dissecting aneurysm arising from A2 segment of anterior cerebral artery (ACA) [1]. The classic presentation of traumatic IAs is hemorrhagic episodes 2-4 weeks after a history of trauma [6].

12. Management

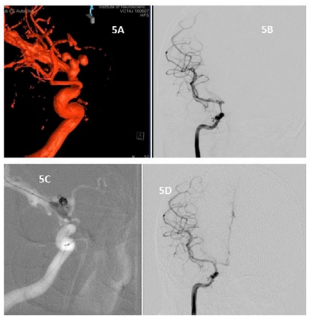
Treatment modality depends upon type of IAs, location, presentation, availability of skilled personnel. The modality with high obliteration and low recurrence rates with low morbidity and mortality should be preferred [10]. The reported favorable outcome for endovascular treatment was 88.3% and for surgical treatment was 82.7% with no statistically significant differences on long term clinical outcome [26]. The overall outcome was found favorable in 84.5% cases in a meta-analysis [26]. In our case, a favorable outcome was observed in 87.5% cases. Dissecting IAs can be treated by either parent artery preserving (reconstructive) or sacrificing (deconstructive) methods [3]. Flow diverting stent is a good option for reconstructive methods which redirect blood away from dissection and press the dissecting flap against the arterial wall [3]. Coils can be used for vessel sacrifice provided collaterals are adequate [3]. In case of infectious IAs both the IAs and the source of infection should be treated [3]. Liquid embolics or glue can be used for ruptured IAs or for occlusion of parent vessel [3]. Traumatic IAs should also be treated by parent vessel occlusion with no attempt for endovascular filling of ruptured pouch6. Surgical options for treating IAs include clipping of aneurysm, ligation of parent vessel, anastomosis and bypass procedures etc [10].



Case 4: Figure 4A, 4B: Pre-operative angiogram revealed saccular aneurysm involving right internal carotid artery bifurcation
Figure 4C: Stent assisted coiling of right internal carotid artery bifurcation aneurysm
Figure 4D: post-operative angiogram following coiling of right internal carotid artery aneurysm with stent insitu.

13. Recurrence and complications

The reported recurrence rate of pediatric IAs is 2.6% annually and of de novo formation or growth is 7.8% which is four times higher than adults [12,27]. That is why, Hetts and colleague considered pediatric IAs to be potentially chronic progressive condition [21]. So, long term follow-up should be scheduled for every IAs regardless of the treatment modality used [10]. Complications may happen but the rate varies among different case series. Vasospasm, infarct, hydrocephalus, meningitis, new focal deficit, other infective complications or death are reported complications [10]. Vasospasm is probably the commonest complication with rates from 9 to 53% [10]. But it is probably more tolerated in children than in adults due to more collateral and great neuroplasticity [8,10]. Overall pediatric population have better favorable outcome than adults [1]. But higher recurrence rate and more de novo formation are important considerations-especially in case of dissecting and fusiform aneurysm [28]. A1 dissecting aneurysm rupture leads to severe hemorrhage resulting in poor prognosis and when re-ruptures has even worse prognosis [29].



Case 5: Figure 5A, 5B: Pre-operative angiogram revealed bilobed aneurysm involving right internal carotid artery bifurcation
Figure 5C: balloon assisted coiling of the aneurysm with coils in the aneurysm
Figure 5D: Follow up angiogram after 3 months

14. Conclusion

Intracranial aneurysms are rare in children but they are getting diagnosed more nowadays. Male are predominantly more affected than female. Usually presents with headache, SAH, seizure or other focal neurological deficit. Besides saccular aneurysms; dissecting and mycotic aneurysms are also common in pediatric population. Endovascular management is an feasible alternative option for treatment of these aneurysms. Though the overall outcome is comparatively better than adult counterpart; regrowth and de novo formation need to be kept in mind.

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