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REVIEW ARTICLE

Ruptured Gint Fusiform Aneurysm in an Infant: Case Report and Review of the Literature

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ABSTRACT

Background: Intracranial aneurysms are exceedingly rare in neonates, accounting for less than 2% of all relevant cases that occur during the first decade of life. More precisely, intracranial aneurysms in children (less than 18 years old) are rarely been recorded, and their epidemiology is poorly investigated. Details about this pathology in this specific population are lacking. Because of the scarcity of this entity along with the peculiar characteristics of this patient group, the treatment of aneurysms of the cerebral circulation of these patients is challenging. The prevalence of Intracranial aneurysms in children is much lower than in adults· apart from that, several differences exist, centered on their etiology, demographic variables, aneurysm location, aneurysm morphological characteristics, clinical presentation, and outcome in pediatric and adult intracranial aneurysms. We describe the case of aneurysmal subarachnoid hemorrhage and extra-axial hemorrhage in an infant, as well as the review of the relevant current literature.

Case description: We report the case of a three months old infant who was admitted to our hospital due to episodes of vomiting, associated with loss of consciousness and an epileptic ictus. An MRI-MRA scan and a digital subtraction angiography were immediately performed, which revealed the presence of a giant fusiform aneurysm near the origin of the right anterior cerebral artery. Because of the location and shape of the aneurysmal sac, the ipsilateral internal carotid artery was totally occluded with detachable coils. At follow-up, the child experienced normal psychomotor development with no motor deficit.

Conclusions: Intracranial aneurysms should be considered in the differential diagnosis of infants and neonates who present with acute raised intracranial pressure. Even though rupture of an intracranial aneurysm in this age group is rare, subarachnoid hemorrhage is the most common mode of presentation. Intracranial aneurysms are frequently larger than 10 mm and located on the middle cerebral artery. The treatment could be surgical or endovascular, depending on the characteristics of the aneurysm. Seizures and cranial nerve involvement are the most common presenting features in children. The most commonly encountered sites of origin for such aneurysms are the posterior circulation, along with the internal carotid artery bifurcation. Current literature and case series suggest that there is increased incidence of giant, posttraumatic, and mycotic aneurysms in children. Based on the most recently published data, the treatment of ruptured and unruptured pediatric aneurysms is related with an increased incidence of an uneventful outcome, which reaches 95% in the current series. Pediatric intracranial aneurysms are more commonly encountered in male patients and have a predilection for the terminal ICA bifurcation.

Abbreviations

ICA: Internal carotid artery
MRI-MRA: Magnetic resonance imaging-Magnetic resonance angiography
GCS: Glascov coma scale
CT: computed tomography
AVM: Arteriovenous malformation
DSA: Digital subtraction angiography
T2 GRE: T2 gradient recalled echo
FSPGR: Fast Spoiled Gradient Echo
ACA: Anterior cerebral artery
MCA: Middle cerebral artery
DWI: Diffusion weighted imaging
3D TOF: 3-Dimensional Time of Flight angiography

INTRODUCTION

Intracranial aneurysms represent an uncommon disease entity in the pediatric population, as they represent 0.5% to 4.6% of all intracranial aneurysms[1,2,3].The vast majority of these cases are encountered among patients that belong to the late adolescence group (0.52 per 100,000 person-years), whereas a significantly lower prevalence is registered within the 0- to 4-year-oldage group (0.06 per 100,000 person-years)[4].The incidence of clinical signs and symptoms seems to be higher in the time period that involves the first 2 years of life[5,6]. More precisely, peak is noticed in the first 6 months and during the second decade of life[7]. Moreover, the incidence of intracranial aneurysms in neonates is exceptionally rare, as only 17 cases have been identified in the last 20years[8].Due to the relative rarity of this disease entity in the pediatric population, scarcity, there is lack of bulk of evidence centered on this pathology in this age group. Apart from that, the pathogenetic factors that are implicated in this target group are not in accordance with those that are related with the adult population. As a consequence, we cannot extract reliable conclusions regarding the offending pathogenetic mechanisms based on adult series.

The predominant location and morphological features, along with the clinical and radiological presentation of pediatric aneurysms are not in accordance with their adult counterparts[9,10]. More precisely, there is a predilection of the internal carotid artery bifurcation and the posterior circulation[11]. Nevertheless, there are several published series that have presented conflicting results when the location, presentation, and clinical outcome of pediatric aneurysms is considered[12,13.14].

Intracranial aneurysms are more frequently encountered in boys when the pediatric population is been considered, and a correlation with collagen mutation diseases such as polycystic kidney disease, fibromuscular dysplasia, Ehlers-Danlos syndrome, or Marfan syndrome seems to be present[15,16,17]. Another difference that exists between the adult and pediatric population is related with the fact that in the pediatric age group a higher prevalence of non-saccular, giant intracranial aneurysms is registered[18,19].Therefore, the treatment of intracranial aneurysms in children and neonates is based on a case-specific basis with a lot of technical issues that have to be addressed. All of these special features seem to be a predisposing factor that leads to an increased morbidity rate. The combination of all of the aforementioned data constitutes the main obstacle to our effort in order to establish a discrete management plan or these patients.

The widespread utilization of new diagnostic and treatment modalities such as magnetic resonance angiography and endovascular intervention techniques will further improve the prognosis of these children. Nevertheless, a delayed diagnosis is a common occurrence in this patient group and, because of that, a high proportion of bleeding or re-bleeding is recorded, which secondarily leads to a poor outcome[12].

CASE DESCRIPTION

We report the case of a three month's old female, generally fit and well, with no known underlying medical condition. She was admitted to our hospital after an ictus of generalized tonic-clonic seizures, accompanied by episodes of vomiting and loss of consciousness. Neurological examination did not reveal any focal neurological deficit and a GCS<8· based on that, the patient was intubated and underwent a CT scan. This revealed the presence of an extra-parenchymal hematoma at the anterior-basal region of the right temporal lobe, in the vicinity of the right temporal pole. Apart from that, there was dense subarachnoid hemorrhage in the anterior basal cisterns and in the interhemispheric fissure. The combination of all of the aforementioned findings necessitated the performance of an MRI-MRA scan. These revealed subarachnoid hemorrhage, with hemorrhagic deposits been noticed in the region of the interpeduncular cistern, left ambient cistern, inter-chiasmatic cistern and fissure, interhemispheric fissure, as well as along the right lateral margin of the tentorium cerebelli. Moreover, this examination verified the existence of an acute extra-axial hemorrhagic collection in the region of the right temporal pole.

We also recognized the existence of a pathological, multi-lobulated mass lesion without

discrete margins, in the vicinity of the right lateral margin of the optic chiasm, at the point of origin of the anterior cerebral artery· this point corresponds to the right margin of the circle of Willis. Regarding this lesion, it extends anterior and superior in relation to the optic chiasm, at the anatomical territory of the gyrus rectus bilaterally, mainly to the right, on either side of the branches of the A2 branches of the anterior cerebral artery. The aforementioned lesion is considered to be inhomogeneous, consisting of a constellation of ovoid lesions, that depict a mixture of low and high signal intensity at T1W images, which simulates with blood products in their acute-subacute phase of deconstruction. Another issue that enhances this concept is related with the recognition of elements that are equivalent with hemosiderin deposition at T2GRE sequence. After the intravenous administration of gadolinium, we recognized the presence of a pathological enhancement of the lesion, whereas the MRA did not reveal the existence of an arteriovenous malformation. We concluded that the described lesion could represent any kind of vascular malformation, more probably a cavernous malformation, which entails hemorrhagic deposits in different time phases. The possibility that this lesion represents a thrombosed AVM is considered a more remote clinical scenario. The MRA recognized the presence of a saccular malformation in the anatomic region of the A1, which most probably stands for an aneurysm, whereas the circle of Willis did not depict any other pathological lesions.

Based on that data, our patient subsequently underwent a DSA of both carotid and vertebral arteries. More precisely, we selectively catheterized both internal carotid arteries and the left vertebral artery and then a 3D rotational angiography with volume rendering was utilized, regarding the arterial branches of the right cerebral hemisphere.

We demonstrated a superior position of the right middle cerebral artery (due to the presence of the extra-axial hematoma), along with moderate vasospasm in the region of the right anterior cerebral artery. This examination revealed the presence of an aneurysm in the territory of the anterior cerebral artery (in the middle of the A1 segment). Its maximum diameter was 3,7 mm and a wide neck was imagined, probably representing a dissecting aneurysm.

We performed a perfusion control of the territory that is nourished from the right ACA, distal to the A2 segment, via the anterior communicating artery and we decided to occlude the aneurysm via detachable platinum coils, along with the relevant proximal A1 segment.

A post-embolization DSA was performed, which verified the occlusion of the aneurysmal sac along with the proximal segment of A1, as well as normal arterial blood supply in the territory of the right cerebral hemisphere.

A post-embolization MRI was performed one month later, which verified the absorption of the extra-axial blood collection, along with the absence of any regions of ischemia in the right cerebral hemisphere, based on DWI. This is in accordance with the fact that no focal neurological deficits were detected and the overall clinical and neurological condition of the patient was excellent. Apart from that, complete occlusion of the A1 segment on the right side is verified with 3D TOF sequence.

A new DSA, three months after the embolization was performed, was revealing no additional imaging findings (verifying the complete occlusion of the aneurysm). A repeat DSA is scheduled for six months after the previous DSA, or earlier, if any clinical suspicion arises.

The main reason that the endovascular approach was preferred over surgery is related with the age of the patient and the goal of the operation. More precisely, as the patient was only a few months old, we considered that it would be beneficial to avoid an operation that could be accompanied with a lot of complications, mainly serious blood loss. Moreover, as our treatment goal was to totally occlude the parent vessel, we decided to use the endovascular route as our first treatment option and in case of failure, our last resort would b an open surgical procedure.

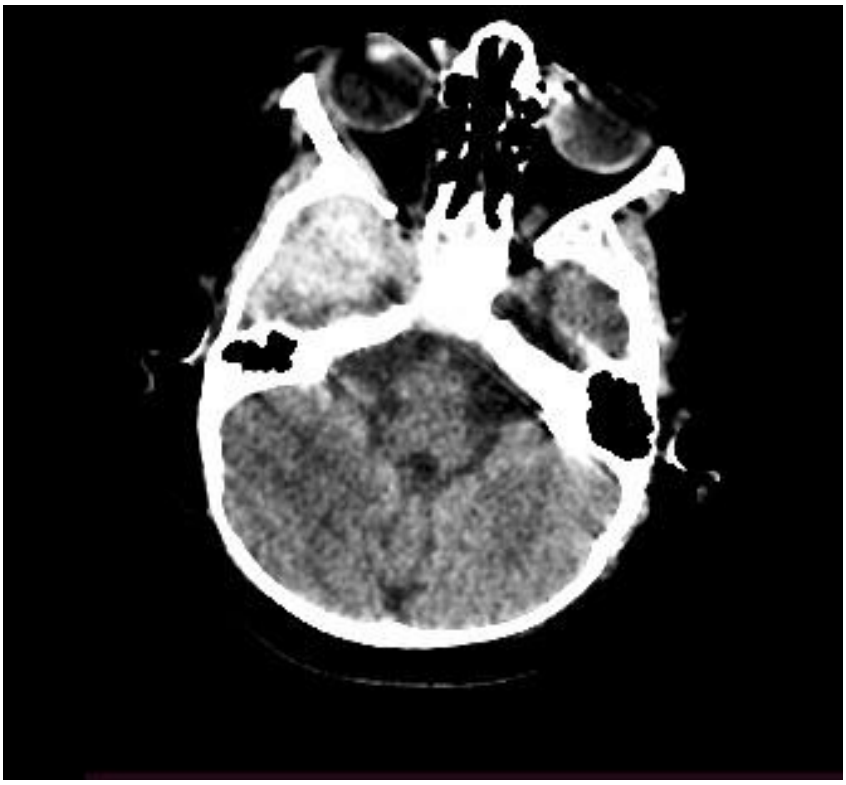

Fig 1. Pre-embolization CT scan, depicting a significant amount of blood clot located in the extra-axial space of the right temporal lobe, in the vicinity of the ipsilateral temporal pole.

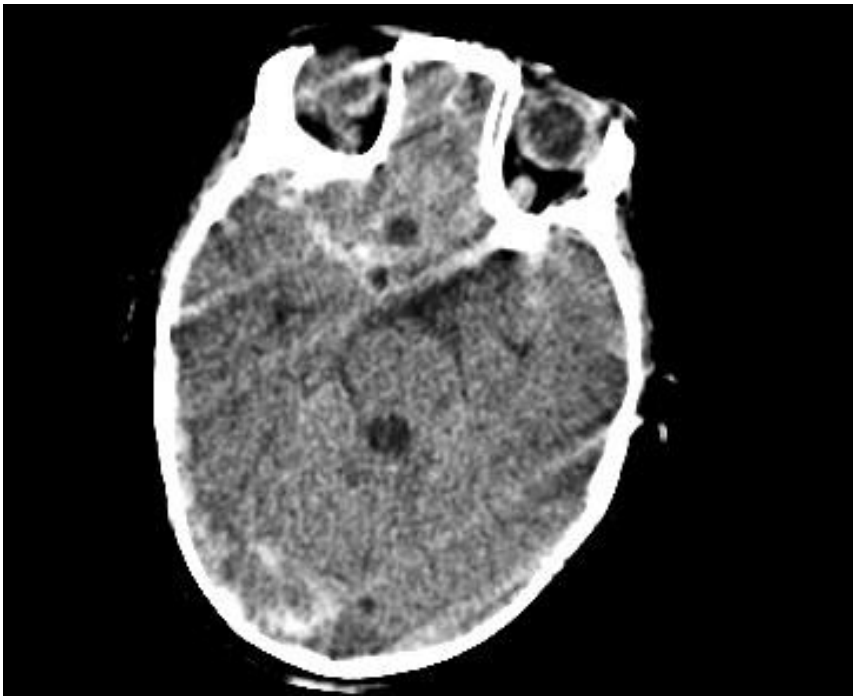

Fig 2. Pre-embolization CT scan, visualizing subarachnoid hemorrhage in the interhemispheric fissure, as well as the basal cisterns.

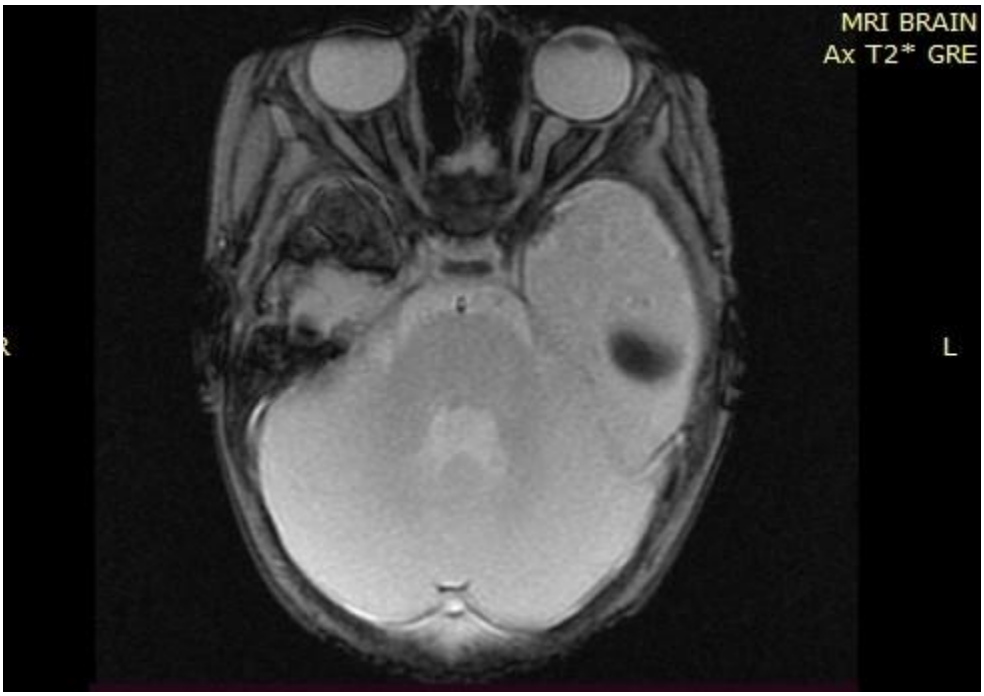

Fig 3. Pre -embolization MRI, axial T2 GRE sequence, verifying the existence of a blood clot in the extra-axial space of the right temporal lobe.

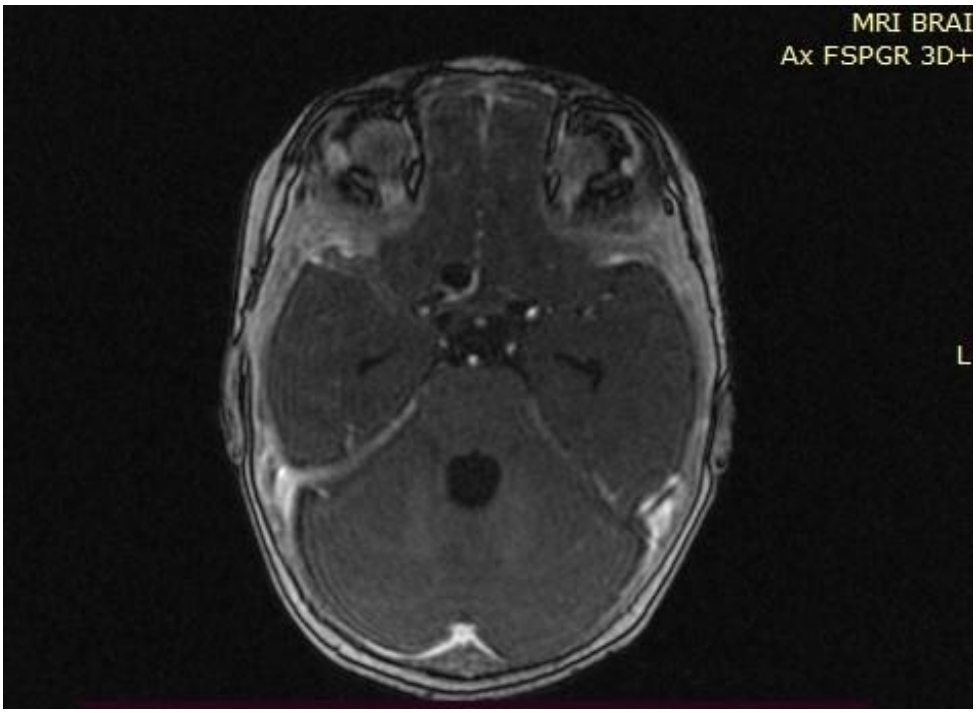

Fig 4. Pre -embolization MRI, axial FSPGR sequence, depicting the subarachnoid hemorrhage in the inter-hemispheric fissure, along with the multi-lobulated mass lesion in the anatomic region of the right anterior cerebral artery.

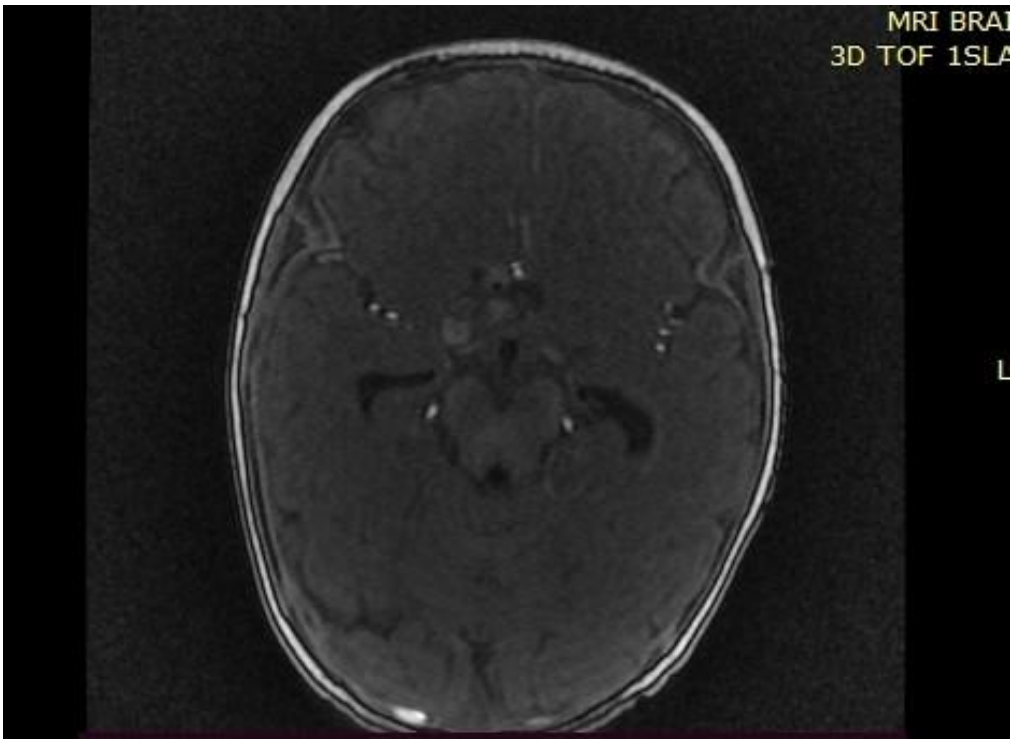

Fig 5. Pre -embolization MRI, 3D TOF, depicting a lesion in the anatomic region of the circle of Willis, which could represent a saccular aneurysm.

Fig 6. Pre-embolization 3D-DSA, depicting a saccular aneurysm in the middle of the A1 segment of the right anterior cerebral artery. It could be regarded as a dissecting type of aneurysm.

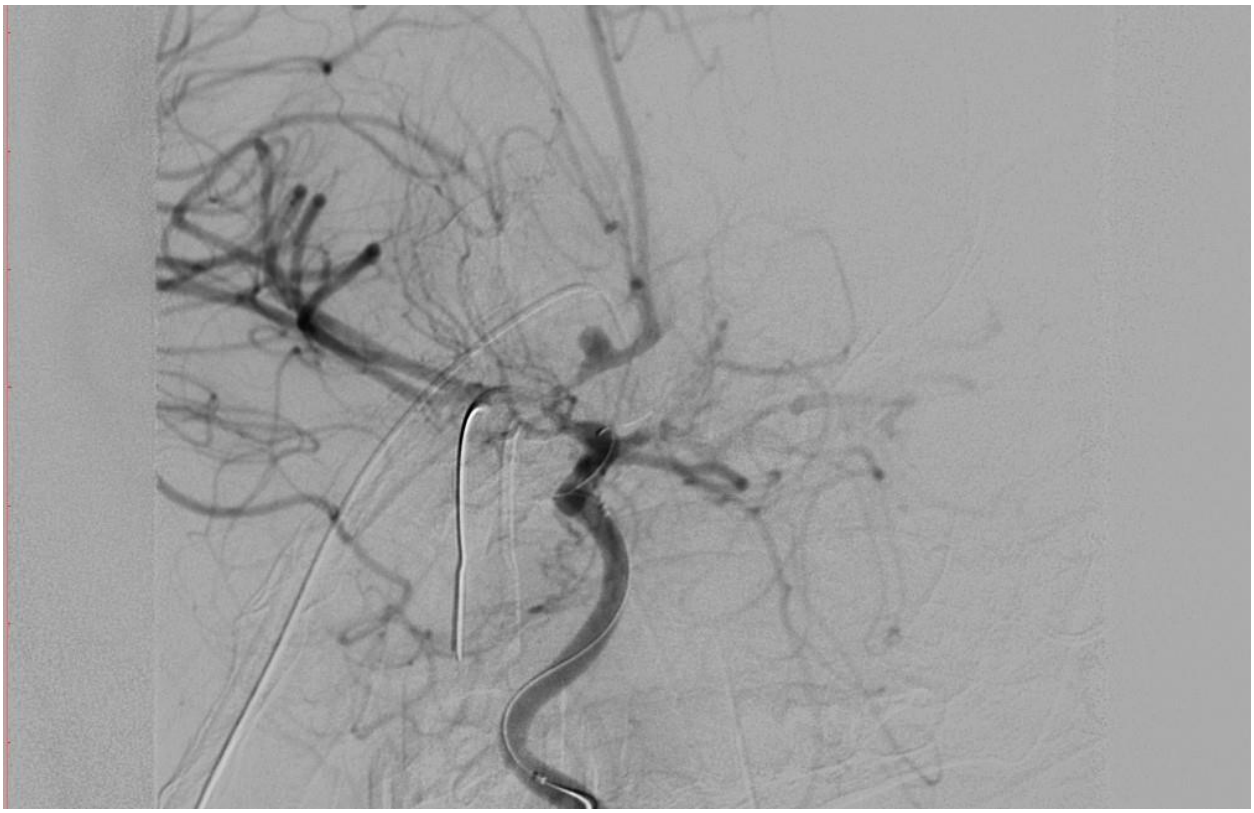

Fig 7. Pre-embolization DSA, lateral view, depicting the site of origin of the offending aneurysm.

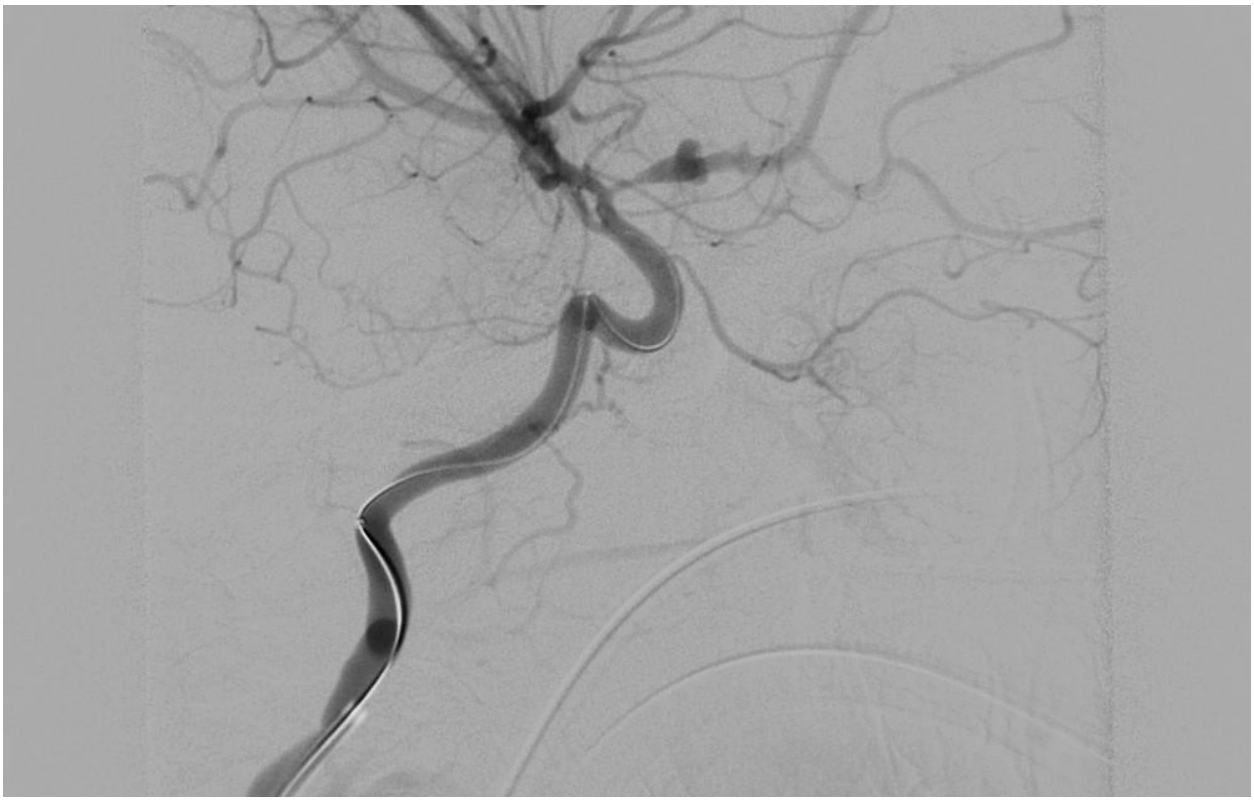

Fig 8. Post-embolization MRI, axial T2 GRE, depicting the complete resolution of the extra-parenchymal blood clot.

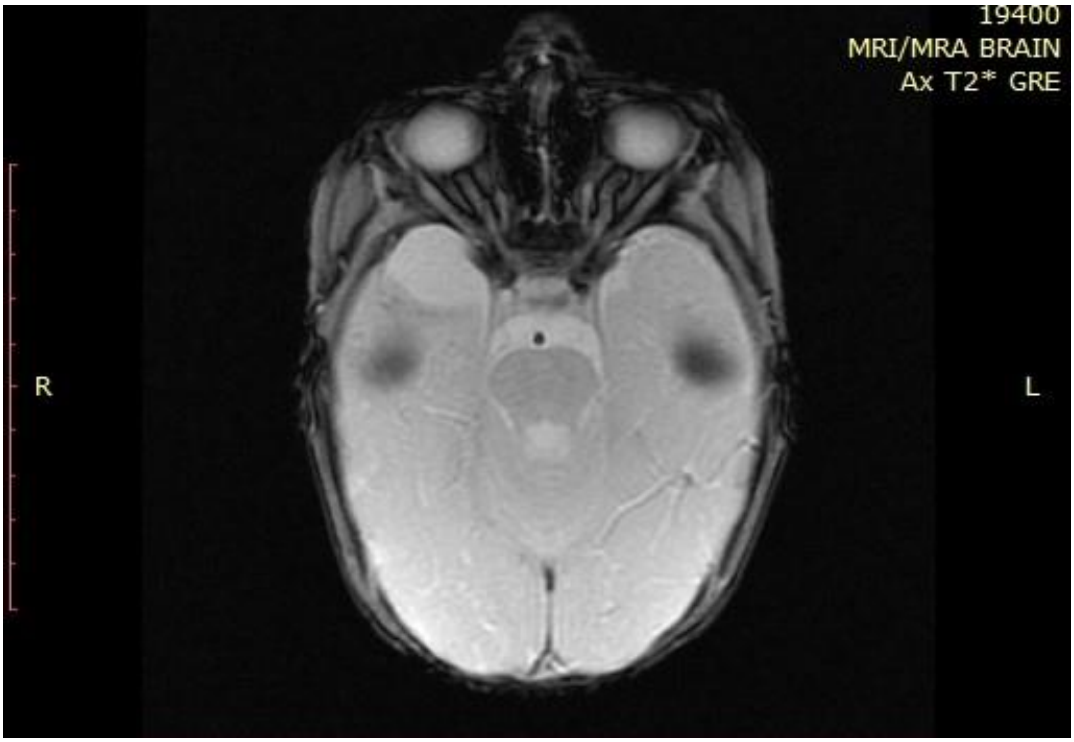

Fig 9. Post-embolization MRI, axial FSPGR, visualizing the occluded aneurysmal nidus, originating from the A1 segment, on the right side.

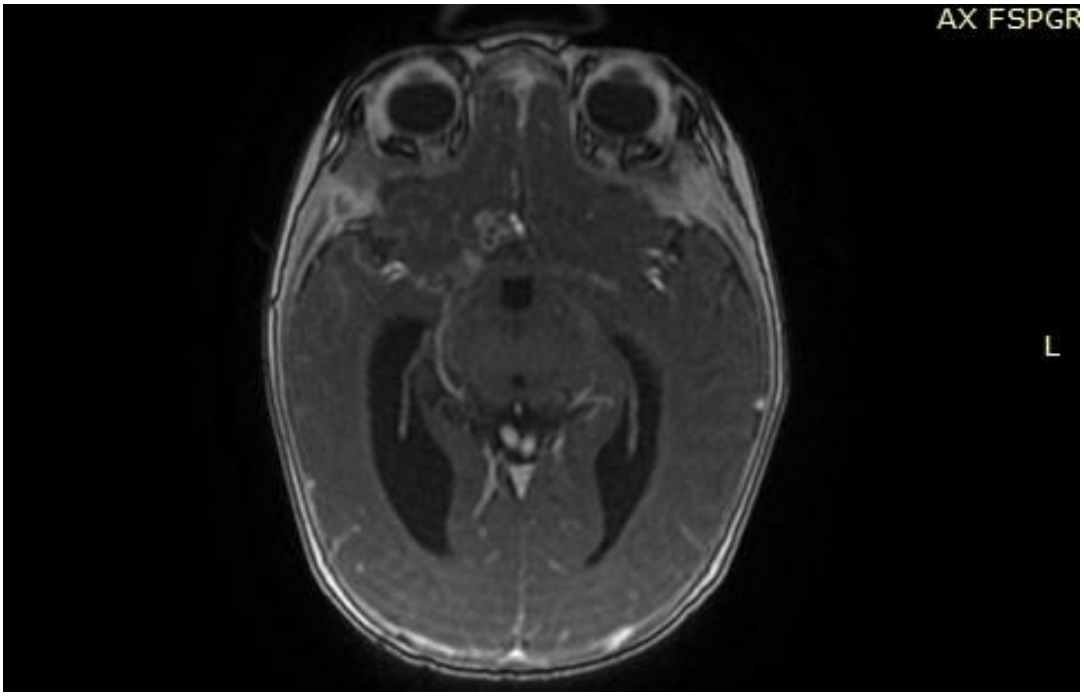

Fig 10. Post-embolization DSA (immediately after the embolization), lateral view, depicting the complete occlusion of the afferent A1 segment, along with the nidus.

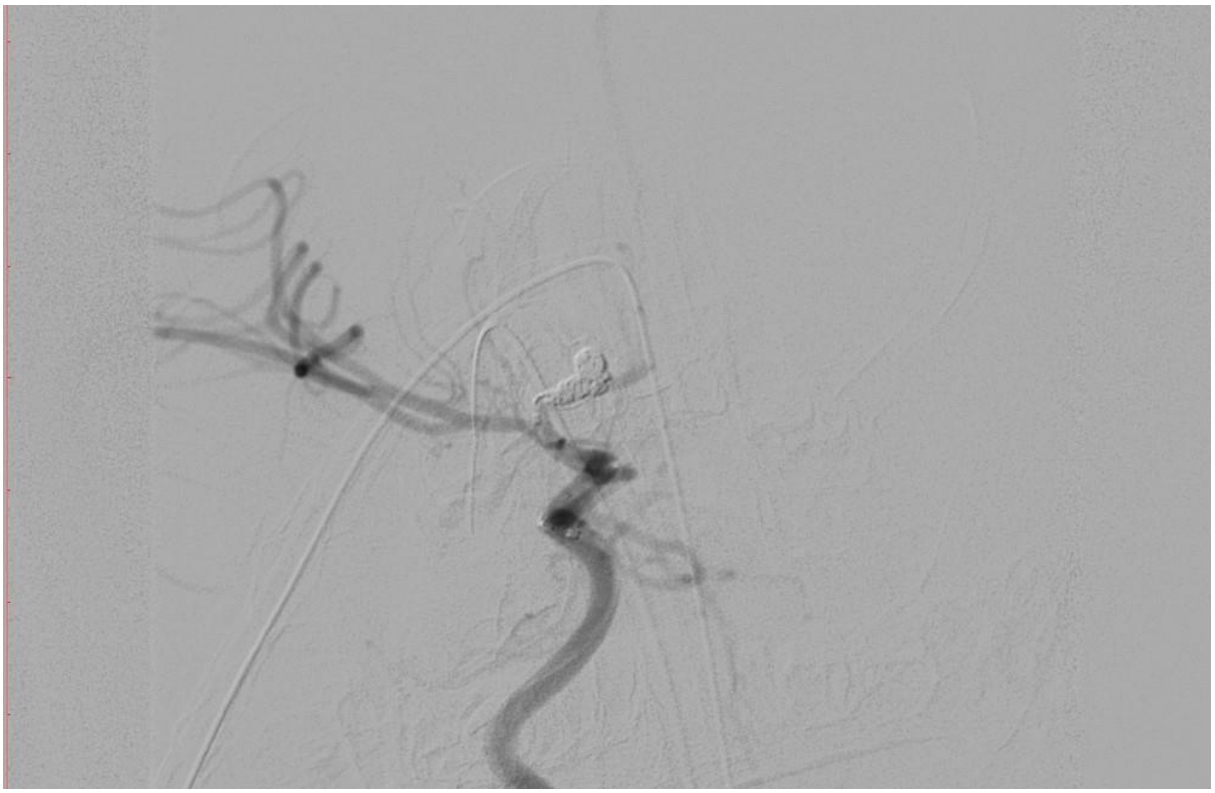

Fig 11. Post-embolization DSA, three months after the endovascular procedure, depicting the same result as the immediate post-embolization DSA.

DISCUSSION

Aneurysms in the pediatric population present with a significantly lower frequency than their counterparts in the adult population. Based on that evidence, the implicated pathophysiological mechanisms along with the relevant treatment outcomes regarding pediatric patients, cannot be extracted with safety, based on data derived from adult patients. This means that there is lack of definitive evidence when the pediatric subpopulation is under investigation. There is bulk of evidence that underlie the existing differentiation when the adult and pediatric aneurysms are compared.

An inherent differentiation that exists between these two subpopulations of patients is based on the different long- life expectancy between them. Children's longer life expectancy is intimately related with the optimum treatment modality that is suggested for these patients. The recommended treatment scheme should be able to achieve high obliteration rates for the aneurysm, along with low recurrence rates, all of them been associated with a low morbidity and mortality rate. Long-term follow-up is mandatory for these patients, as there is a life-long probability of aneurysm recurrence, along with de novo aneurysm formation. All of these parameters render the establishment of definitive, widely accepted treatment recommendations for this subset of population even more difficult. This is the reason why such a consensus still is lacking from the available literature.

Demographics

Intracranial aneurysms in the pediatric population are rare, accounting for less than 5 % of all intracranial aneurysms[20,21,22]. A relevant review was based on children who were ageless than one year and were harboring intracranial aneurysms. According to them, only 131 of such patients fulfilled those criteria[23]. Based on literature data, the male to female ratio varies from 1:1.2 to 2.8:[12,21,22,24, 25.]

Clinical presentation

The constellation of clinical features regarding the pediatric population cannot be described based on the adult counterparts. According to a large series[20], headache constitutes the most commonly encountered complain of the involved patients, a report that was verified by other researchers[22,24]. Loss of consciousness was the first clinical finding in 27% of patients in the previously reported research[20], whereas a greater percentage of patients had a medical history of an ictal loss of consciousness in other studies[22,24]. More precisely, seizure was the presenting feature in 21% of patients in the previously reported series, whereas the existing literature is exhibiting a wide spectrum

of relevant results[22,26]. Krishna et al[22]mentioned that an epileptic ictus could be considered as the presenting feature in a much more significant portion of pediatric patients, compared to in adults (36 vs. 17 %, p value <0.05). Limb weakness at the time of presentation is a common clinical feature, although the reported prevalence varies from 9%[25] to41%[22]. Cranial nerve involvement is a commonly recorded accompanying clinical feature, with literature data reporting a variation from 26 %[24] to 50 %[22].

Another finding that is almost universally accompanying all cases of aneurysmal rupture in the adult population is subarachnoid hemorrhage. The reported incidence of SAH in literature varies from 58 to 91 %[3,22]. The discrepancy in the incidence of SAH in the pediatric population compared with the adults has been proposed to be related with the high incidence of mycotic, traumatic, and giant aneurysms in pediatric patients[25].

Another clinical point that differentiates the pediatric from the adult population is based on the clinical observation that children have a tendency to present with a better clinical picture after aneurysm rupture, compared to their adult counterparts[21]. The proposed underlying pathophysiology under this condition may be related to factors such as fewer comorbidities, and a greater tendency to refer cerebrovascular cases to tertiary centers[27].

Radiological findings

The prevalence of intracranial, aneurysm-related, hemorrhage that is ICH and IVH, was varying among several relevant 26 %series. More precisely, Garget al[20] have recorded an incidence rate in the range of 26%, whereas Sharma et al.[25] reported hemorrhage in 36 % of their participants. Hydrocephalus complicated 28%of patients, according to a large single center experience, which is comparable with the relevant results that were derived from other case series studies (29 to36%)[22,25]. One of these studies mentioned that hemorrhage and hydrocephalus were more commonly encountered in children, compared to their adult counterparts· nevertheless, this difference was not statistically significant[22].

Another point that deserves special mention is related with the size of the aneurysms in this age group. There are several reports which state that aneurysms in the pediatric age group tend to be large in size. Krishna et al[22] reported that the prevalence of giant aneurysms is more than double in children, when they are compared with their adult counterparts, whereas an approximate estimation of the reported incidence of giant aneurysms in children ranges from 0 to 54 %[20,28,29,30,31].

Another point of differentiation between the two subgroup of patients is related with the relative incidence of multiple aneurysms in a single patient. Literature data state that the recorded incidence of multiple aneurysms in children varies from 0 to16 %[3,15,22,32], Krishna et al. reported that multiple aneurysms are less commonly encountered in children than in adults[22].

Another important point that differentiates pediatric and adult patients is centered on the existing differences in the aneurysm location[27,30,32]. Whereas Garg[3]stated that anterior circulation aneurysms constitute the vast majority of such aneurysms, accounting for 82 % of all aneurysms, Jianet al[27] concluded that the incidence of posterior circulation aneurysms in children was considerably larger, compared to the relevant percentage that was recorded in adults[32]. When the anterior cerebral circulation was investigated, the commonest points of involvement, regarding ICA aneurysms, were the ICA bifurcation (23%) followed by the cavernous segment (16%)[3]. This is in contrast to the adult population, where aneurysms located in that territory constitute25–39 % of all aneurysms[3,33].

Certain aneurysms have been grouped under the term complex aneurysms, because they exhibit unusual characteristics, when their morphological features, location, or etiology are analyzed[21]. This term underlies giant, mycotic, traumatic, or multiple aneurysms, dissecting aneurysms, as well as those with unusual locations such as distal MCA or intra-cavernous ICA. The relative percentage ofpatients that are recorded in the literature as harboring complex aneurysms varies from 27 % to 91%[22,24,33,34].

Management

Surgical intervention techniques that were involved in pediatric patients include clipping of aneurysm and ligation of proximal vessel. As the prevalence of complex aneurysms in this patient population is relatively increased, a high incidence of utilization of innovative procedures such as anastomosis and bypass procedures have been mentioned in literature[12,21,35]. A surgical method was considered as the procedure of choice in a percentage of patients that varies from 33 to 100% in various series[3]. Pediatric intracranial aneurysms share in common several peculiar features that differentiate the treatment algorithm from their adult counterparts.

As we have already mentioned, a significant percentage of these aneurysms are characterized as complex· because of that, the appropriate

treatment protocol requires a more sophisticated treatment plan. This includes procedures that are not restricted to surgical aneurysm clipping or endovascular coiling, and include vascular anastomosis and bypass procedures. Moreover, the life expectancy of children is far more extended compared to that of adults. Because of that, the expected incidence of aneurysm recurrence and new aneurysm formation is undoubtedly increased and this is intimately related with the time course of these patients. This observation is supported by a relevant study[26].All these data support the concept that the proposed treatment modality for a patient that belongs to the pediatric age group should be able to provide long term safety and efficacy.

It is widely accepted that the currently preferred method for the management of intracranial aneurysms in the adult population is the endovascular approach. A large relevant study[34] compared the long-term results of the microsurgical and endovascular approach when they have been used for the treatment of a children group of patients. They concluded that an obliteration rate in the range of 82 and 94% was related with the patient groups that underwent an endovascular and microsurgical treatment, respectively.

Aneurysm location

Another feature that differentiates adult from pediatric patients is related with the fact that, according to a large relevant study[22], the occurrence of multiple aneurysms is more common in children, compared with their adult counterparts. Apart from that, pediatric and adult patients are differentiated when the location of the aneurysm is taken into consideration[15,27,32], with a predilection of the anterior circulation for aneurysms that are related with pediatric patients[20]. However, there are reports[20,27] which have mentioned that children more frequently harbor aneurysms of the posterior circulation, with a relative frequency that is greater when compared with the corresponding frequency, when their adult counterpart is investigated.

A discrete subpopulation of aneurysms has been overall characterized as complex aneurysms, based on their morphological features, location, oretiology[20,21]. Under this term are included giant, mycotic, traumatic, or multiple aneurysms, dissecting aneurysms, as well as a small subset of aneurysms that favor an unusual location. These include distal MCA or intra-cavernous ICA. There is a wide range of the estimated proportion of pediatric patients that from 27 % to 91 % in the literature[22,33,34].

Follow-up

Based on the fact that patients that belong to the pediatric age group share in common a long-life expectancy, they are intimately associated with an increased risk of aneurysm recurrence, along with de novo aneurysm formation. The combination of all these features makes mandatory the adoption of long-term follow-up for children that are treated for intracranial aneurysms, no matter what was the treatment modality used. According to a large relevant study[27], if microsurgical clipping was the selected treatment modality and there is no residual aneurysm on post-operative DSA, the suggested follow-up study includes a MRA on a yearly basis. The frequency of long-term surveillance can eventually be decreased to once every 5 years. As far as the aneurysms that are treated via an endovascular route are under consideration, the authors have proposed that a DSA should be done 6 months after the treatment. If this examination does not reveal recanalization, the recommended protocol for their follow-up suggests an annual MRA, which could be gradually replaced by an MRA every 5 years.

Conclusion

Health care professionals that treat pediatric patients should always bear in mind the fact that intracranial aneurysms in the pediatric population share in common several unique characteristics that differentiate their clinical presentation, location, treatment options and long-term follow-up from their adult counterparts. Among these differences, we would like to underline the fact that aneurysms are rarely encountered in children less than 5 years of age and that they occur in males are recognized with twice the frequency in relation to females.

Another peculiarity that is inherently related to this group of patients is associated with the fact that seizures and cranial nerve involvement are more frequently encountered as the presenting features in children· besides, this was the presenting feature of our patient. There is a predilection for the posterior circulation, when pediatric population aneurysms are considered, as well as for the bifurcation of the ICA. Finally, giant, post-traumatic, and mycotic aneurysms in children are more commonly encountered than in adults and endovascular and microsurgical treatment has been adopted as treatment options, without any profound evidence that support the superiority of one of these techniques over the other.

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