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Case Report

Brain AVM with Accompanying Venous Aneurysm with Intracerebral and Intraventricular Hemorrhage

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Abstract

Background: Arteriovenous malformations (AVM's) and intracranial aneurysm are collectively the most common causes of spontaneous subarachnoid and intracerebral and/or intraventricular hemorrhage. In children, saccular aneurysms can be attendant to the AVM in 29% of children with AVMs. The majority of aneurysms are arterial in location (37%), with a similar percentage of intranidal (30%) and venous (33%) locations.

Case Presentation: A 13 year's old male was admitted to our hospital due to gradually deteriorating level of consciousness. CT and MRI revealed intra-parenchymal and intraventricular hemorrhage. Work up included a DSA which revealed an AVM with an associated venous aneurysm. Endovascular embolization of the AVM was executed and the postoperative course of the patient was benign.

Conclusions: Contrary to many other pediatric centers, our clinical practice has been to perform urgent cerebral angiography in children with suspected AVMs to identify and potentially treat AVM-associated aneurysms. We have done this because of our hypothesis that AVM- associated aneurysms have a higher risk of re-hemorrhage than isolated AVMs.

Keywords: *Arteriovenous Malformation; Associated Venous Aneurysm; Rupture; Embolization*

Abbreviations

CT: Computed Tomography; MRI: Magnetic Resonance Imaging; DSA: Digital Subtraction Angiography; AVM: Arteriovenous Malformation; AV: Arteriovenous; T2 W: T2 Weighted; T1W: T1 Weighted; T2 GRE: Gradient Echo Sequence; FLAIR: Fluid-Attenuated Inversion Recovery

Introduction

Arteriovenous malformations are one of the major causes of stroke in children [1,2]. Because intracerebral hemorrhage is the most devastating as well as the most common clinical presentation (46% - 87%) in children with AVMs; there is a strong interest in establishing treatment strategies that could prevent or reduce both the initial hemorrhage and possible re-bleeding. Even though AVMs are rare in kids, estimated to represent 3% of all AVMs [3,4], they tend to rupture more frequently than in adults [4,5].

An enormous diversity of brain vascular malformations occurs in children. These include vein of Galen malformations, dural AV fistulas, non-Galenic pial AV fistulas, and nidal arteriovenous malformations (AVMs) [6]. AVMs are defined by a group of vessels with an abnormal low-resistance connection between arteries and veins occurring in a focal geographic area of the brain parenchyma - the nidus.

Aneurysms separate and distinct from the circle of Willis are commonly found located adjacent to or within AVMs, and are typically classified as either arterial, intranidal, or venous in location [1]. The nature of these AVM-associated aneurysms has been partially investigated in adults.

Although the data are mixed, the preponderance of data suggests that the presence of an aneurysm may be an independent risk factor for hemorrhage in adults with AVMs. For this reason, most adults with ruptured AVMs undergo prompt cerebral angiography with endovascular treatment of AVM associated aneurysms. In children, however, conventional cerebral angiography and treatment are often performed in a delayed fashion, in part because the nature of AVM associated aneurysms is generally unknown. Because pediatric and adult aneurysms may have different causes, extrapolation of data from adult studies may not be applicable. In this study, we describe our experience regarding the clinical behavior and treatment of AVM-associated aneurysm in a child with AVM.

Case Report

A thirteen year's old male was admitted to the emergency department of our hospital due to progressive loss of consciousness with a rapid clinical deterioration. Upon admission, CT and MRI were performed, which revealed intraparenchymal hemorrhage in the region of the splenium of the corpus callosum with accompanying intraventricular hemorrhage in the left lateral ventricle (Figure 1a-1j).

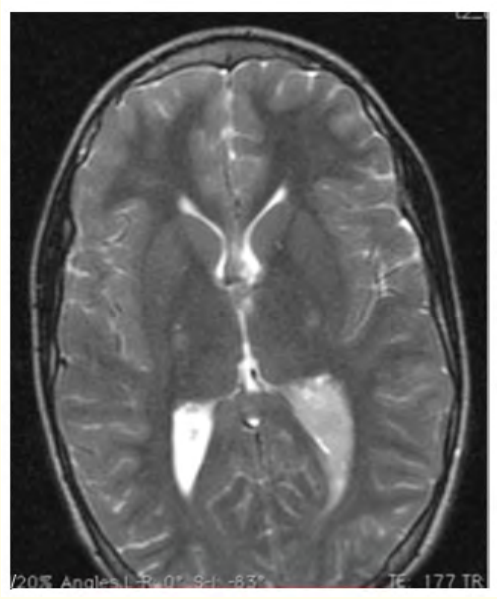

Figure 1a

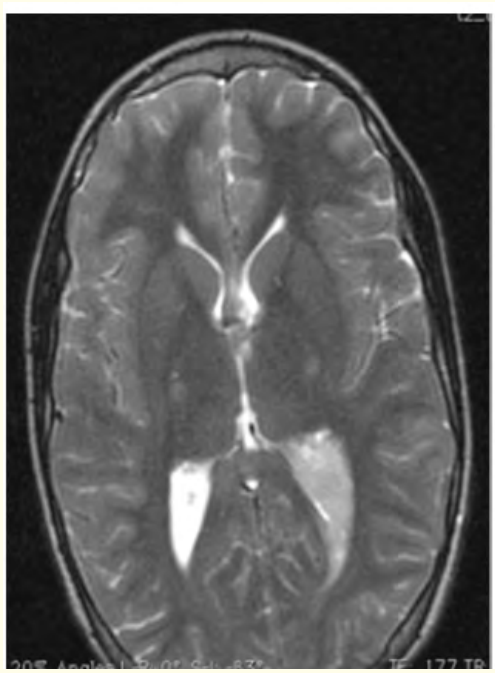

Figure 1b

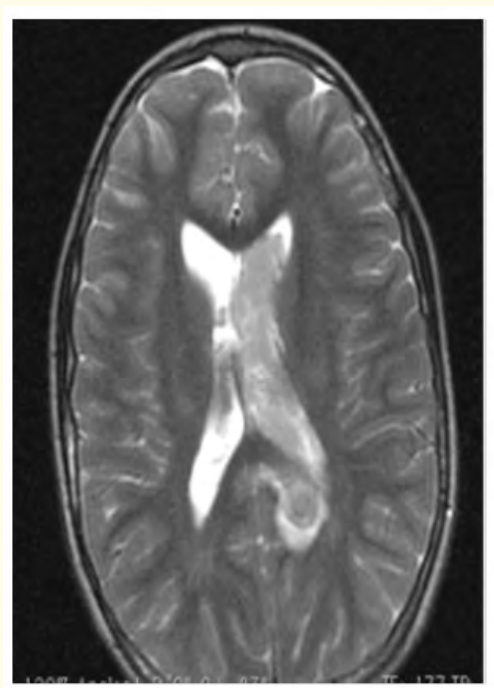

Figure 1c

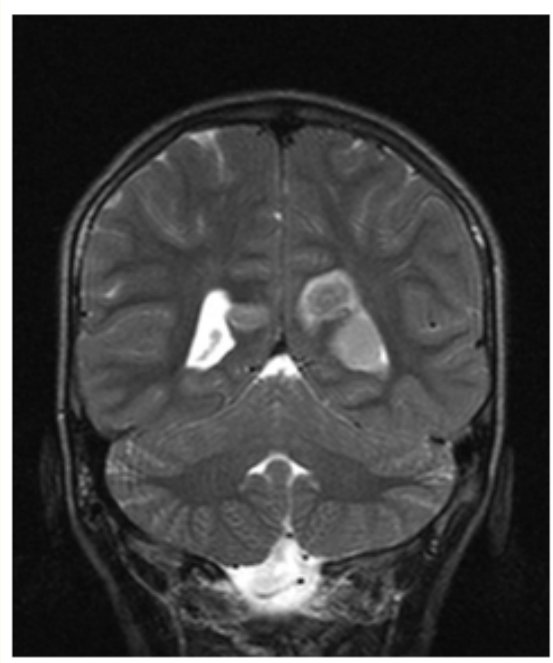

Figure 1d

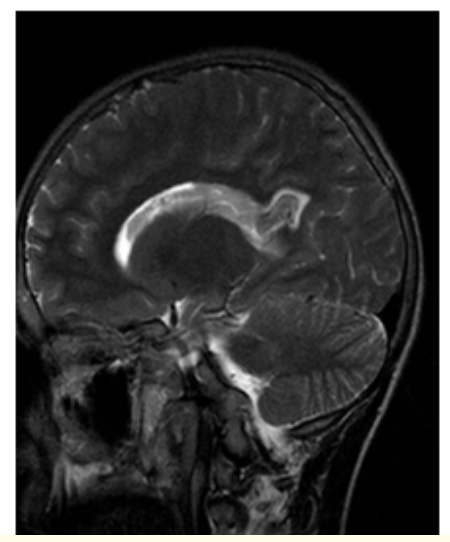

Figure 1e

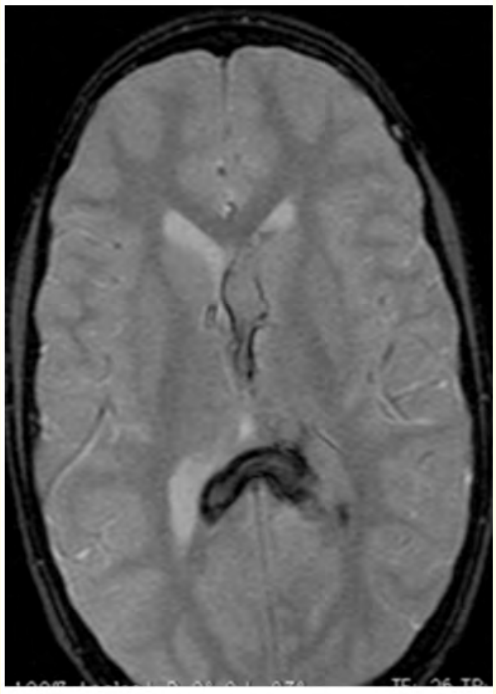

Figure 1f

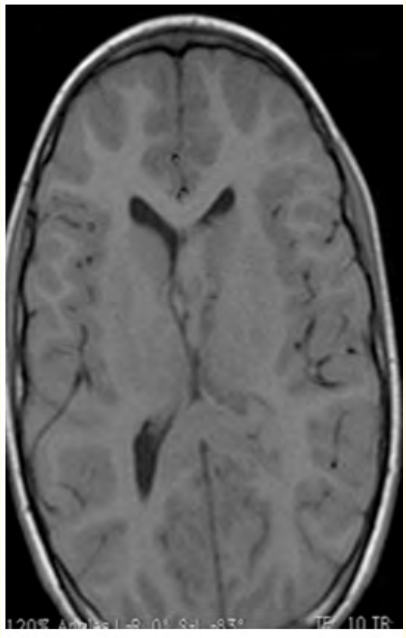

Figure 1g

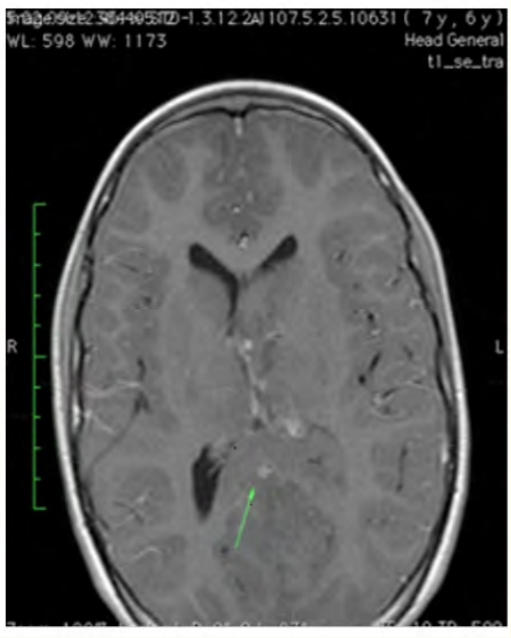

Figure 1h

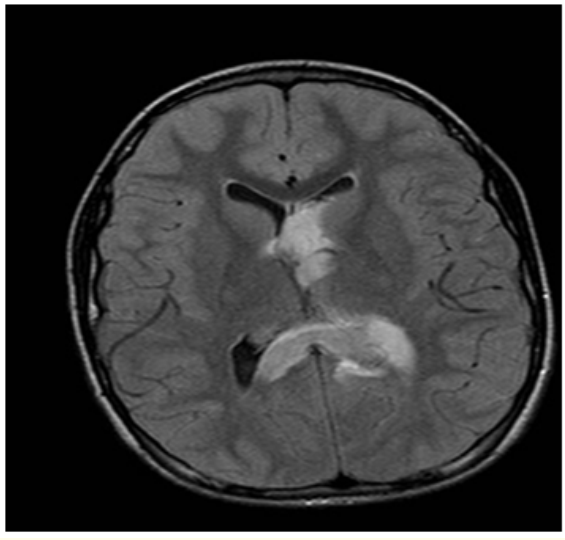

Figure 1i

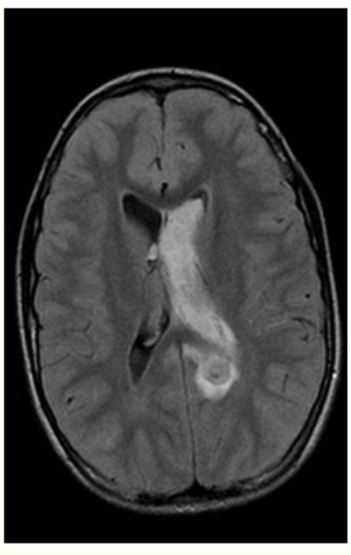

Figure 1j

Figure 1a-1j: *Initial MRI scan, T2 W, T2*GRE, T1W before and after gadolinium enhancement and FLAIR, revealing intra-parenchymal hemorrhage in the anatomic territory of the splenium of the corpus callosum and the isthmus of the cingulate gyrus. An accompanying intraventricular hemorrhage in the left lateral and third ventricle is visualized. After gadolinium administration, there is moderate contrast enhancement. The long arrow at figure 1h depicts the location of the nidus in the territory of the splenium of the corpus callosum.*

DSA was performed, which revealed an underlying AVM with an accompanying venous aneurysm in the vicinity of the splenium of the corpus callosum. Arterial feeders were originating from the left side of the posterior cerebral circulation, whereas the draining venous channels were terminating to the left transverse venous sinus (Figure 2a-2d).

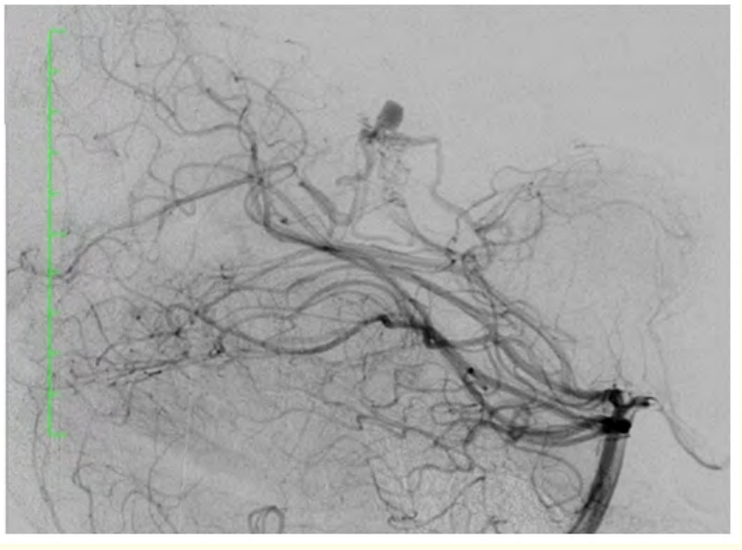

Figure 2a

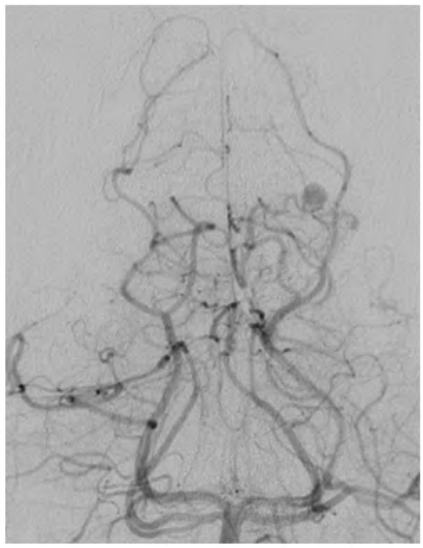

Figure 2b

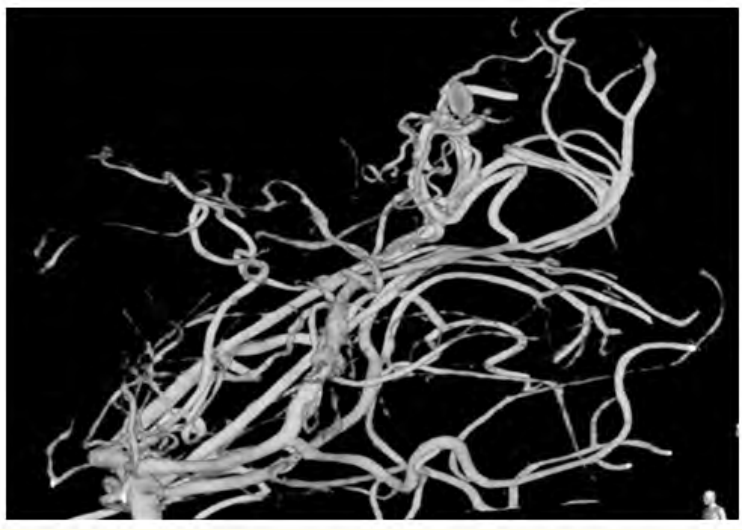

Figure 2c

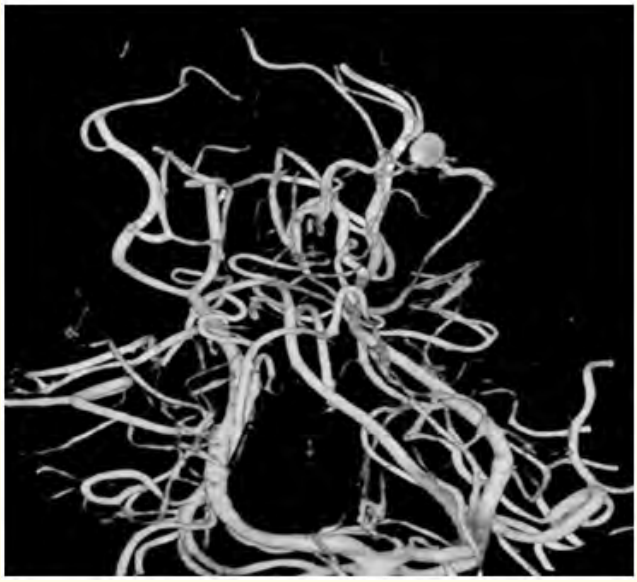

Figure 2d

Figure 2a-2d: *Initial DSA of posterior cerebral circulation, revealing the underlying AVM with the associated venous aneurysm. Arterial feeders of the nidus are branches of the left posterior cerebral artery.*

Subsequently, the patient underwent trans-arterial embolization of the AVM through elective catheterization of the left posterior cerebral artery.

After embolization, a small residual remnant of the nidus of the AVM remained with delayed and slow opacification of the draining vein of the malformation (Figure 3a-3c).

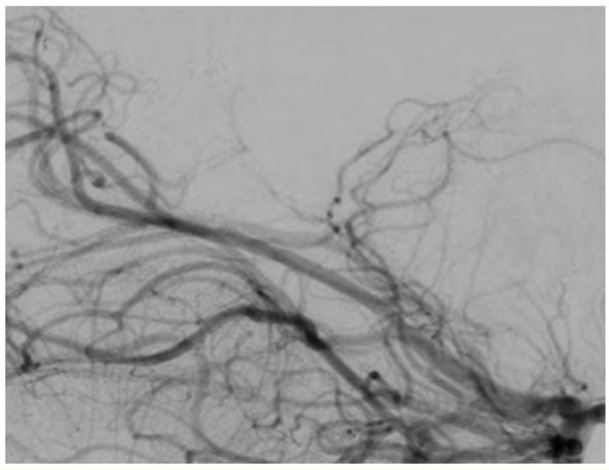

Figure 3a

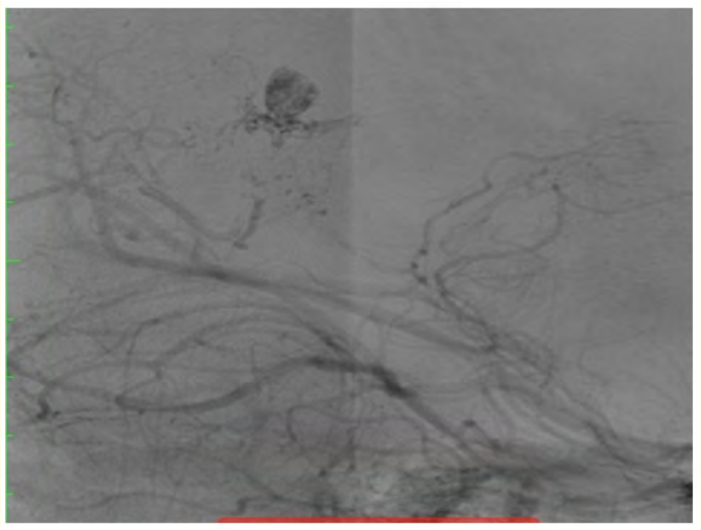

Figure 3b

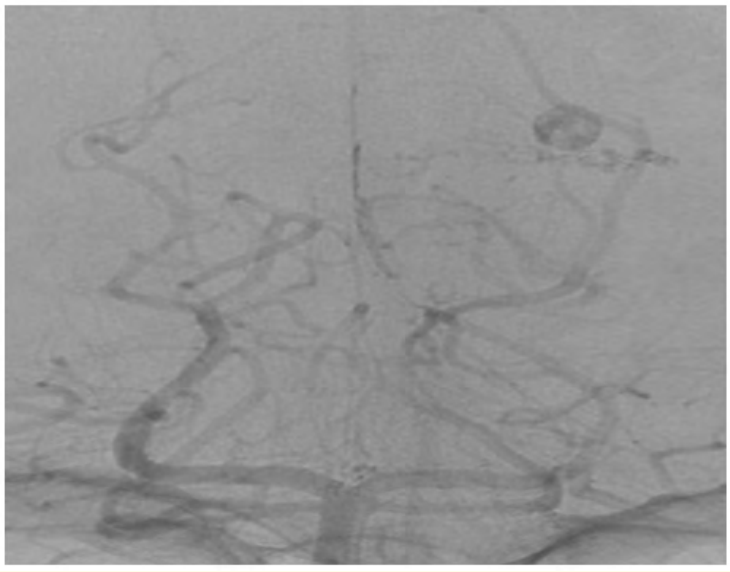

Figure 3c

Figure 3a-3c: *Elective DSA of the posterior cerebral circulation, after selective catheterization of the left posterior cerebral artery. It reveals a small residual portion of the nidus remaining patent and thus opacified, with concurrent slow, delayed filling of the draining vein.*

After the embolization procedure, a CT scan was performed, which verified the embolization result and that no adverse consequences, especially hemorrhage and infarction in the territory, were incurred (Figure 3d). The patient's clinical course was uneventful, with gradual recovery of the neurological and cognitive function.

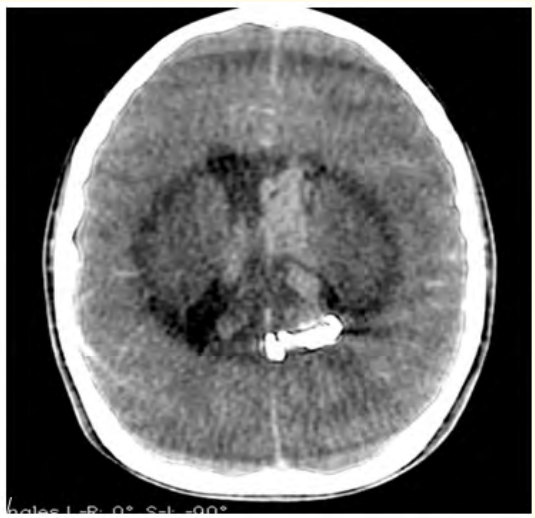

Figure 3d

Figure 3d: *CT scan performed immediately after the embolization procedure. It delineates the location of the occluded nidus of the AVM as a hyperdense area. No adverse effects, such as hemorrhage or infarction, are noted.*

A follow-up DSA was performed four months after the ictus, which revealed the existence of an early draining vein in the vicinity of the known AVM, without residual opacification of the nidus of the AVM (Figure 4a-4b).

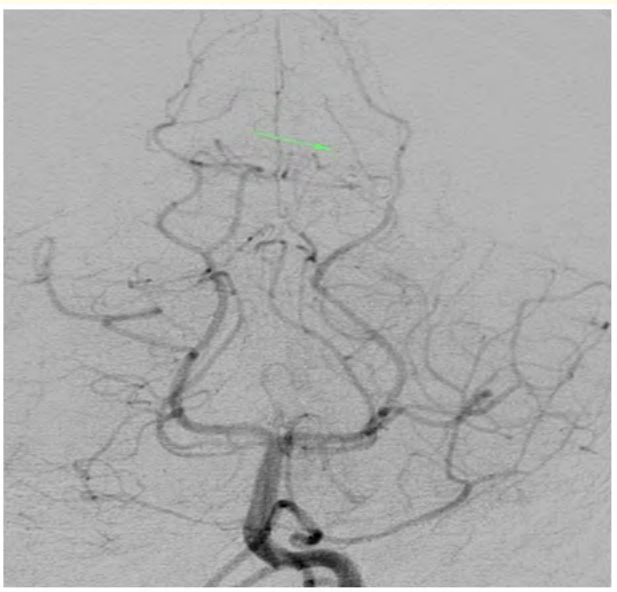

Figure 4a

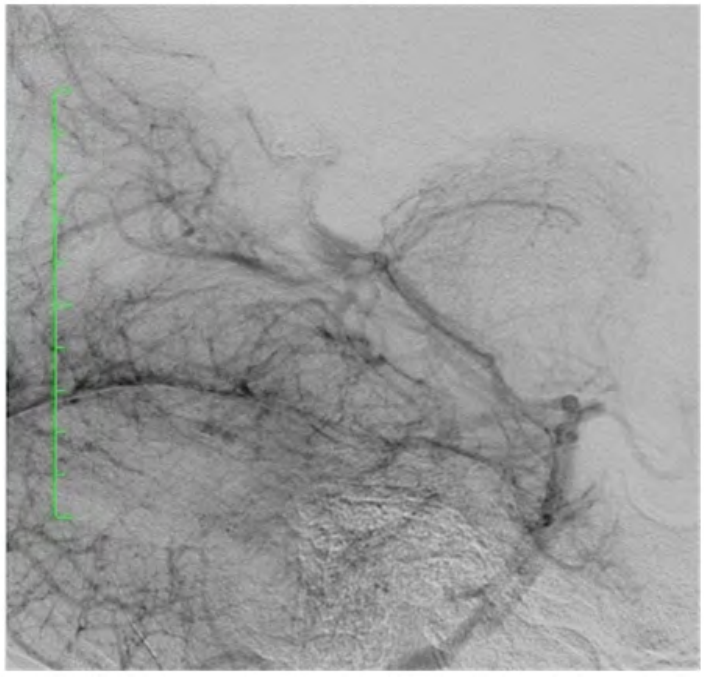

Figure 4b

Figure 4a-4b: *DSA performed four months after the embolization procedure, revealing complete occlusion of the nidus, which is not opacified. It only depicts the existence of an early draining vein.*

Results and Discussion

In a large study reporting the overall incidence of AVM associated aneurysms in children, as well as their association with hemorrhage, an AVM-associated aneurysm was found in 29% of children with AVMs [1]. The majority of aneurysms were arterial in location (37%), with a similar percentage of intranidal (30%) and venous (33%) locations. The presence of an aneurysm could not be predicted based on patient age, sex, or location of the AVM.

AVM-associated aneurysms in venous and intra-nidal locations (48% of patients) did not appear to be associated with hemorrhage when separated from arterial aneurysms.

AVMs and their draining veins were often located deep within the brain in children, raising the possibility that centrally-located AVMs may arise earlier in development or be more likely to come to clinical attention early in life than more peripherally located AVMs.

Regarding clinical status, most studies that demonstrate that children with AVMs overall improve clinically over time [1,7]. Comparison of clinical status at initial presentation, discharge, and most recent follow-up all suggested that while children with AVM associated aneurysms have worse neurological status at discharge, there was no significant difference between children with isolated AVMs and those with AVM-associated aneurysms at long-term follow-up.

It has been routine clinical practice to perform urgent cerebral angiography in children with suspected AVMs to identify and potentially treat AVM-associated aneurysms [1]. This is based on the hypothesis that AVM-associated aneurysms have a higher risk of rehemorrhage than isolated AVMs.

Regarding treatment of pediatric AVM's, endovascular embolization is feasible and safe in treating pediatric AVMs; it will continue to evolve and improve and should be incorporated into the treatment paradigm for adjunctive treatment in the pediatric population [3].

Conclusion

Some researchers advocate that microsurgical resection remains the gold standard for the treatment of all accessible pediatric AVMs, especially in cases where urgent intervention is needed such as in acute intracranial hemorrhage [3]. We illustrate a case with a not easily accessible AVM with an acute cerebral hemorrhage in an eloquent brain region that could not be evacuated operatively, so we did not prefer this strategy.

On the contrary, other researchers have stated that early angiography with endovascular treatment of arterial-based aneurysms in children with AVMs may be indicated [1] this strategy was the one followed by us, with excellent clinical outcome.

The optimal management for pediatric AVMs remains controversial. Lifelong risks of bleeding and potential deficits are relatively high compared to the adult population. The technology for the management of these lesions is still evolving. A multidisciplinary approach using multimodality therapy if needed has been proved to be beneficial in approaching these lesions in all age groups [3].

Conflict of Interest

I declare that no financial interest or any conflict of interest exists.

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