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CASE REPORT

An incidental pure arterial malformation in a child: case report and review of the literature

Meena Thatikunta[1] · Natasha V. Raman[1] · Kristin N. Zieles[1] · Katrina Ducis[1] · Andrew Jea[1]

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Abstract

Purpose Pure arterial malformations of the brain are rare vascular lesions.
Methods We present a 10-year-old healthy boy who presented with an incidental finding of pure arterial malformation.
Results Our case seems to represent the second description of pure arterial malformation discovered incidentally in a child.
Conclusion We review the clinical presentation, angiographic findings, and management of our case in the context of other reported pediatric cases.

Keywords Arterial malformation · Angiography · Pediatric neurosurgery · Endovascular

Introduction

Recently, greater attention has been garnered on rare vascular malformations of the brain —"pure arterial malformations"— in both the adult and pediatric age groups [1–9]. Moreover, pure arterial malformations are thought to be associated with a benign natural history and should be treated conservatively. At times, these malformations may be mistaken for arteriovenous malformations (AVMs), arteriovenous fistulas (AVFs), or cerebral saccular aneurysms [2–5].

✉ Andrew Jea
ajea@iuhealth.org

Meena Thatikunta
Meena.thatikunta@gmail.com

Natasha V. Raman
nraman@iuhealth.org

Kristin N. Zieles
kzieles@iuhealth.org

Katrina Ducis
kducis@iuhealth.org

[1] Section of Pediatric Neurosurgery, Department of Neurological Surgery, Riley Hospital for Children, Indiana University School of Medicine, 705 Riley Hospital Drive, Suite 1601, Indianapolis, IN 46202, USA

Incidental presentation of pure arterial malformation in otherwise healthy children has been less frequently described [9]. In this study, we describe the clinical course of one child over 12 months of follow-up.

Case report

History

Our patient is a 10-year-old boy who presented after a concussion suffered during a hockey game. He had no relevant past medical history. A noncontrast CT of the head (Fig. 1) suggests abnormal cerebral vasculature. The patient was otherwise asymptomatic.

Examination

Patient displayed a normal neurological examination without focal deficits. There was no pronator drift.

Diagnostic studies

Advanced imaging with CT angiogram (CTA) of the head (Fig. 2) revealed a tortuosity of vessels in the left internal carotid artery segments. Patient then underwent a diagnostic cerebral angiogram (Fig. 3). There was no aneurysm, AV

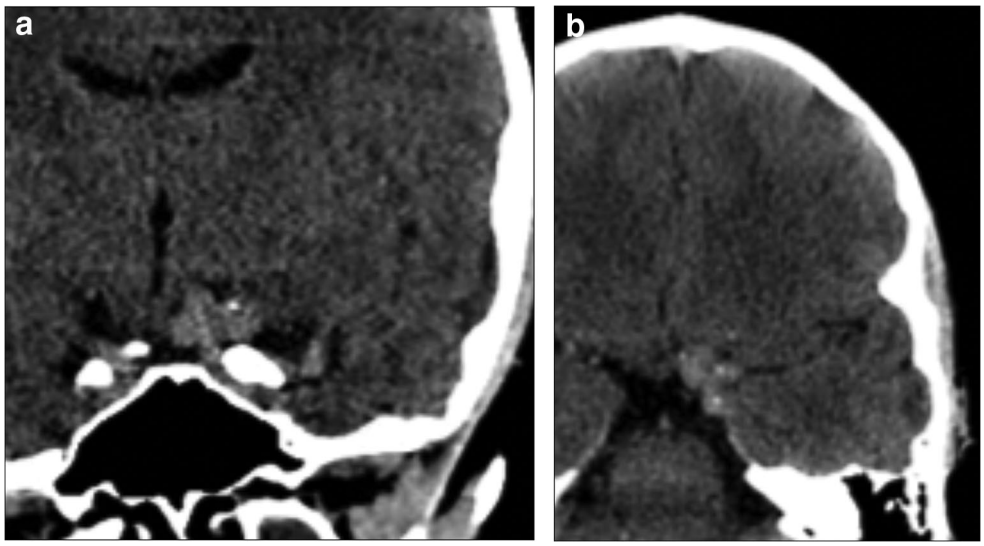

Fig. 1 CT head. **a** Coronal and **b** axial noncontrast head CT shows a hyperdense mass with eggshell calcifications in the region of the supraclinoidal segments of the internal carotid artery, suggestive of a vascular malformation

fistula, or early venous phase drainage associated with this malformation.

Clinical course

The patient did well throughout the year of follow-up and at 1 year after the procedure. He remained asymptomatic. There was no growth or change in the morphological appearance of the lesion based on follow-up CTA. Because our patient was asymptomatic with no changed based on noninvasive imaging, the risks of invasive conventional angiography did not seem warranted. He has also returned to playing hockey.

Discussion

Our case was consistent with the previously published incidental case of pure arterial malformation in a child (Table 1) [9].

In one review [9], the authors found that the most common locations for pure arterial malformations were the ACAs and PCoA-PCA complex, as in our patient.

Catheter angiography is an important component to distinguish these entities. Differentiating between AVMs and AVFs, and pure arterial malformations is straightforward as there is no arteriovenous shunting in pure arterial malformations [4, 5].

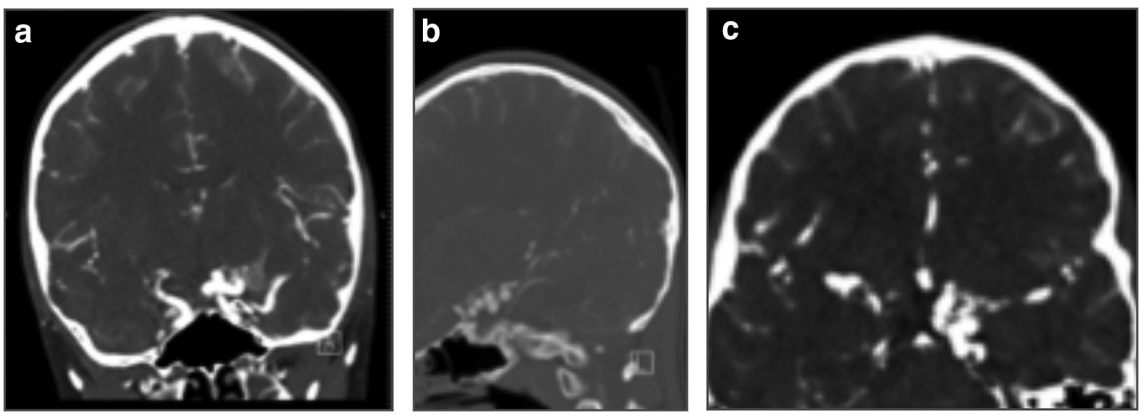

Fig. 2 CTA head. **a** Coronal, **b** sagittal, and **c** axial projections of the CTA head confirm a vascular malformation of the left cervical internal carotid artery (ICA), distal ICA, anterior cerebral artery, and fetal posterior cerebral artery

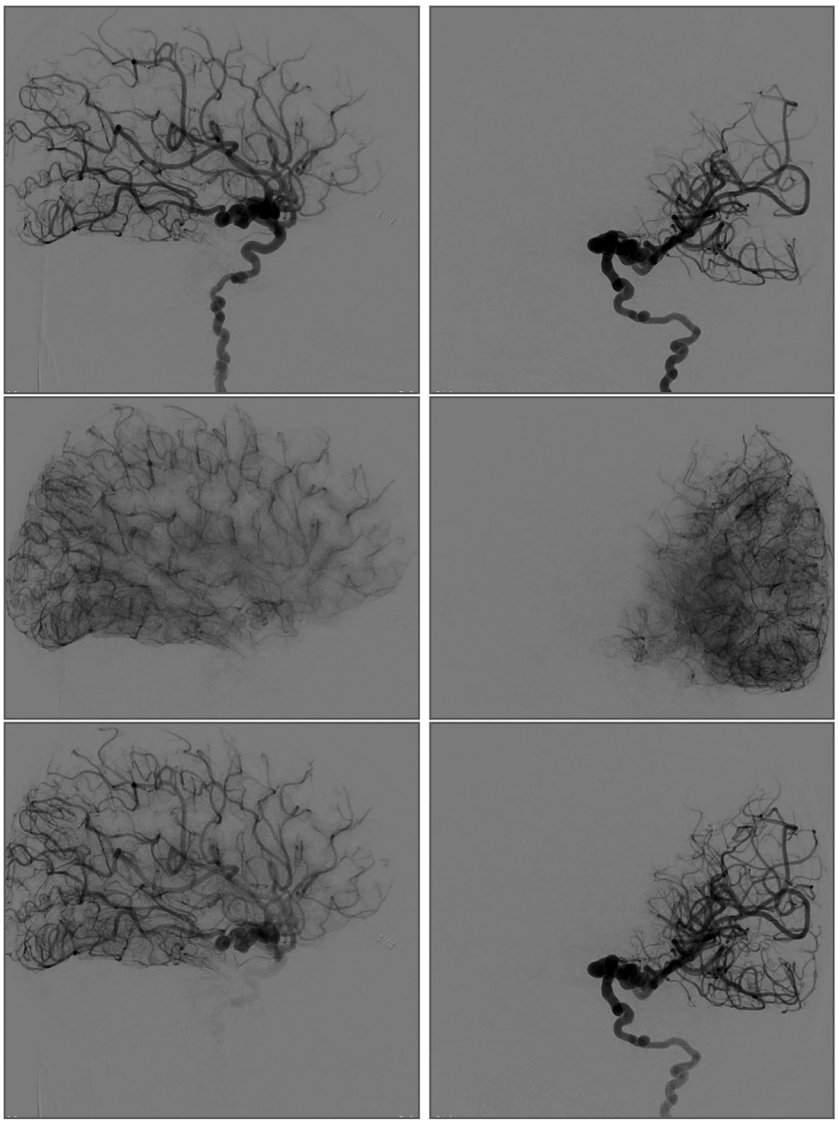

Fig. 3 Digital subtraction angiography of the left internal carotid artery (ICA) injection demonstrates vessel tortuosity in the cervical and distal ICA segments and a fetal posterior cerebral artery

The etiology of pure arterial malformations is unknown. Some hypotheses include an embryologic defect or insult during development of the intracranial arterial circulation resulting in dysplasia, segmental arterial vulnerability from an acquired insult such as viral infection or genetic mutation later in life, or a chronic healed traumatic dissection of a major intracranial artery [9].

Conservative management as a mainstay of treatment for pure arterial malformations is consistent with case reports and small case series published in the literature [2–9].

Conclusions

Pure arterial malformations are a rare type of brain vascular malformation in the pediatric patient population. Most reported cases have been associated with diseases such as viral infection, PHACE syndrome, and moyamoya disease. Incidental presentation in children is even less common. Diagnosis of these lesions may be challenging; however, catheter angiography may help ascertain the diagnosis of pure arterial malformation. Invasive treatment is generally not indicated. Management consists of surveillance with serial imaging such as MRA.

Table 1 Incidental (*italic type*) and symptomatic cases of pediatric pure arterial malformation

Case report/series	Age	Gender	Symptoms	Location	Diagnostic imaging	Treatment	Follow-up
Brijikji et al. *2018*	*11*	*M*	*Incidental*	*Left PCoA*	*Coiled*	*None*	*No change at 1 month*
Sorenson et al. *2018*	*17*	*F*	*Migraine*	*Right PICA*	*Coiled loops with fat elements*	*None*	*No change at 8 years*
Brijikji et al. 2018	10	F	Headache	Left supraclinoid ICA, PCoA, PCA	Multilobulated aneurysm of supraclinoid ICA; partially calcified, dilated, tortuous PCoA and PCA	Coiling of aneurysm associated with PAM	No change at 72 months
Brijikji et al. 2018	17	M	Headache	Right SCA	Dilated and coiled	None	No change at 26 months
Sacks & Lindenburg 1969	2	M	Viral encephalitis	Bilateral A2	Dilated and coiled	None	None
Laterna et al. 2014	1	M	Infarct from moyamoya	Left PCoA and PCA	Dilated, coiled, ipsilateral moyamoya	None	None
Vanslambrouk et al. 2000	5	M	Right hemiparesis	Left ICA, PCoA, PCA, MCA, and SCA causing brainstem compression	Coiled, dilated	None	None
Baccin et al. 2007	4	F	PHACE syndrome, right hemiparesis	Left supraclinoid ICA and PCoA causing infarct	Coiled, very dilated	None	No change at 16 months
Baccin et al. 2007	1	F	PHACE syndrome, fever, and hypotonia	Left supraclinoid ICA, PCoA, PCA, R supraclinoid ICA	Coiled, very dilated	None	None
Metry et al. 2001	1	F	PHACE syndrome	Left MCA and supraclinoid ICA	Coiled, dilated	None	None
Yamada et al. 1985	17	F	Nausea, vomiting	Left supraclinoid ICA, MCA, and ACA	Coiled, ectatic, calcified	None	None

PICA, posterior inferior cerebellar artery; *PCoA*, posterior communicating artery; *ICA*, internal carotid artery; *MCA*, middle cerebral artery; *ACA*, anterior cerebral artery

Compliance with ethical standards

Conflict of interest The authors have no conflicts of interests to report.

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