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Case Report

Rare case of giant pediatric cavernous angioma of the temporal lobe: A case report and review of the literature

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ABSTRACT

Background: Giant cavernous malformations of the central nervous system are quite rare. They are more common in children and may be misdiagnosed as other intracranial neoplasms. Here, we presented a very rare giant cavernous angioma mimicking a neoplastic temporal lobe lesion in an 18-month-old male.

Case Description: An 18-month-old male presented with two initial seizures. Although the clinical examination was normal, the computed tomography (CT) scan showed a large left temporal mass (66 mm diameter) exerting significant mass effect and midline shift. The brain magnetic resonance (MR) imaging demonstrated a large left temporal heterogeneously enhancing lesion with significant perilesional edema and mass effect. The patient underwent gross total removal of the lesion that proved to be an intracranial cavernous angioma. Postoperatively, he did well, exhibiting no residual neurological deficit, and has remained lesion and seizure-free.

Conclusion: This and 12 other cases in the literature focus on intracranial cavernous angiomas that could have been readily misdiagnosed as tumors. It confirms why obtaining appropriate preoperative MR and CT studies, followed by surgical intervention, is essential to confirm the correct underlying pathology and appropriately and optimally treat the patient.

Keywords: Cavernomas, Cavernous angioma, Giant cavernous malformation, Pediatric patients

INTRODUCTION

Intracranial cavernous malformations (CMs) are benign low-flow vascular lesions that make up to 10–25% of all vascular malformations, 70–80% are supratentorial.[1]

Most CMs are small and remain asymptomatic for long periods of time. Occasionally, supratentorial CMs will precipitate the new onset of seizures and headaches, while infratentorial CMs more typically lead to acute/progressive neurological deficits.[5,6]

Few giant CMs (GCMs) have been reported in the literature and may mimic intracranial neoplasms or other vascular malformations. Here, we present an 18-month-old male with a GCM and reviewed 12 other cases in the literature.

CASE REPORT

An 18-month-old male presented with the new onset of two seizures. Although the clinical examination was normal, both the computed tomography (CT) and magnetic resonance (MR)

studies showed a large left temporal lesion with severe mass effect and midline shift. The enhanced MR further demonstrated heterogeneous enhancement (lesion >66 mm) and perilesional edema [Figure 1].

Surgery

The left temporal craniotomy revealed a large mass with distinct margins from the surrounding tissues accompanied by gliosis. The pathology was consistent with a cavernous angioma. Although the patient did well immediately postoperatively, 3 days later, he developed meningitis that was treated effectively with 3 weeks of antibiotics. Postoperatively, the patient remained neurologically intact without seizures. The subsequent CT studies 3 months later confirmed no recurrent lesion [Figure 2].

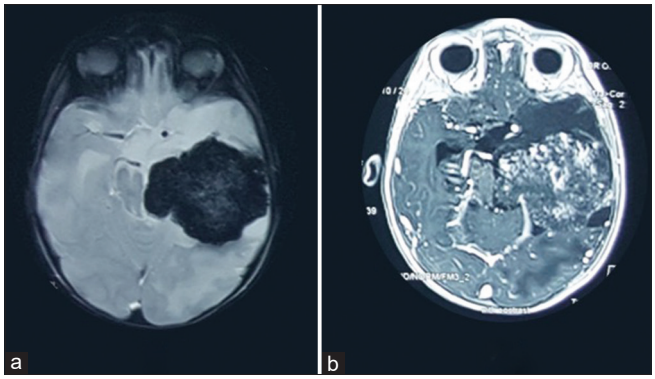

Figure 1: Brain magnetic resonance imaging (MRI) of a left temporal giant cavernous angioma (a) MRI gradient spin echo weighted imaging showed a lesion with a diameter over 66 mm a hypo signal on gradient spin echo weighted imaging (b) brain MRI T1-weighted imaging with gadolinium injection showed a heterogeneous enhancement of a left temporal lesion with brain edema.

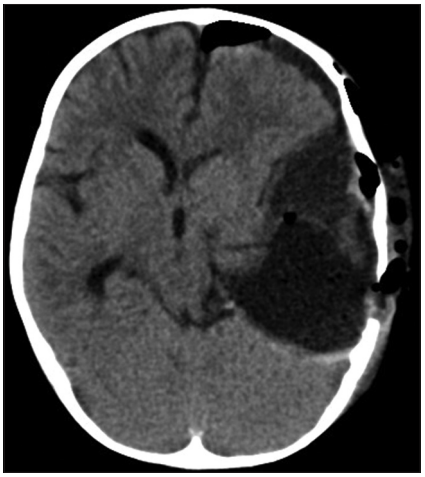

Figure 2: Postoperative brain computed tomography scan showing a complete removal of the cavernous angioma.

DISCUSSION

History

In 2004, Lawton *et al*. defined a giant cavernous angioma as a lesion with a diameter over 60 mm.[10]

Jhawar *et al*.[6] reviewed 16 cases of giant cavernomas in all age groups but ranging in size from 4 cm and 6 cm. All lesions were reported as solitary and most frequently were found in the frontal and frontoparietal regions [Table 1]. One case cited a right temporo-parieto-occipital giant cavernomas: our case study uniquely involved the temporal region alone.

Frequency

Cavernous angiomas are vascular benign malformations of the central nervous system.[1] The prevalence is about 0.5%.[6] They represent 5–10% of all intracranial vascular malformations. They are more common in adults in the second to fifth decades and are most frequently found in the supratentorial white matter[1] (e.g., parietal lobe and thalamus).[6]

Lesion size and clinical presentation

Although these lesions are typically 9–20 mm, giant lesions also have been reported in the literature. In both children and adults, these lesions may first present with focal or generalized seizures followed by intracerebral bleeding and deficits reflecting lesion location. Rarely, pediatric cases of giant cavernomas have been reported in the literature [Table 1]. In 1984, Khosla *et al*. first reported on a pediatric giant cavernoma; since then, there have been just 12 more cases.[9]

CT and MR appearance

Typically, the MR imaging (MRI) appearance of a giant cavernous angioma is a heterogeneous "popcorn-like" mass with/without cysts reflecting various states of degradation of blood.[12] The hypointense halo on T2-weighted images is due to hemosiderin deposits. In the present case, the left temporal lesion was isodense on CT scan with heterogeneous enhancement. The MRI showed a 66 mm, heterogeneous lesion that showed a hypointense halo on T2-weighted MRI imaging and did not enhance with gadolinium. This appears to be the 13th case of a giant cavernoma [Table 1]. The mechanism of lesion enlargement was likely attributed to repeated bleeding episodes.[13,14]

Misdiagnosis of giant cavernous angioma

Giant cavernous angioma can be misdiagnosed on CT/MRI, as they mimic other intracranial neoplasms. Therefore, other

Table 1: Reported pediatric case of giant cavernous angioma in the literature.

S. No.	Author	Year	Age	Sex	Size (cm)	Location
1	Khosla *et al.*[9]	1984	3 years	Male	>6	Left frontoparietal
2	Kawagishi *et al.*[7]	1993	11 months	Male	8	Right pineal and trigone
3	de Andrade *et al.*	2002	7 years	Female	12×14	Left frontoparietal
4	de Andrade *et al.*[4]	2002	7 years	Female	>6	Left frontoparietal
5	Chicani *et al.*[3]	2003	15 years	Male	7×5	Left parieto-occipital
6	Lawton *et al.*[10]	2004	12 years	Male	13×7×7	2/3 anterior right hemisphere
7	van Lindert *et al.*[15]	2007	3 months	Female	6–7	Left frontotemporal and paraventricular
8	Avci *et al.*[2]	2007	14 years	Male	4.2×3.5	Left frontoparietal
9	Gezen *et al.*[5]	2008	10 months	Male	6×4×4.5	Left parietal
10	Agrawal *et al.*[1]	2012	14 years	Male	4.2×4.5	Left temporoparietal
11	Agrawal *et al.*[1]	2012	10 years	Female	5×4	Left frontal
12	Ozsoy *et al.*[11]	2017	10 years	Male	8×7.5×7	-
13	Our reported case	2017	18 months	Male	6.6	Left temporal

differential diagnosis include gliomas, oligodendrogliomas, pilocytic astrocytoma, or with hemorrhagic metastasis.[1,13]

Surgery

The operation of choice is gross total excision, but this depends on lesion location. Stereotactic radiosurgery might be considered for deep sited lesions, but correlate with poor outcomes.[8] The efficiency of radiosurgery for cavernous angioma remains uncertain.

Pathology

Giant cavernoma consists of a large ectatic endothelium-lined variable sizes vascular channels without mural muscular or elastic fibers embedded within a matrix of collagenous tissue. There are no neuronal cells or other components. Further, it is surrounded by hemosiderin deposits, gliosis, and sometimes calcification or thrombosis.[6]

CONCLUSION

Pediatric giant cavernous angioma is a rare intracranial lesion that may be best diagnosed with MR/CT. The best outcomes correlate with surgical excision, but maybe, limited by eloquent tumor location.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms.

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Conflicts of interest

There are no conflicts of interest.

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