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Case Report

Unusual cause of binocular diplopia: Cavernous sinus hemangioma [☆]

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ABSTRACT

Cavernous hemangiomas are rare account representing 5%-13% of the intracranial vascular malformations and occur in approximately 0.5%-1% of the population. We report the case of 34-years-old woman, having a medical history of seizure, admitted for headache with binocular diplopia. The radiology investigation and operatory piece has shown an association of cavernous sinus hemangioma and frontal focal cortical dysplasia. This study highlights clinical, radiological and therapeutic features of this entity.

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Introduction

Cavernous hemangiomas are vascular malformations comprised of large venous spaces with thrombosis and hemorrhage. Intracranial localization is rare, representing 5%-13% of the intracranial vascular malformations and occur in approximately 0.5%-1% of the population [1]. Clinical presentation varies from epilepsy, headache, and cranial nerve palsies. CT and MR imaging plays an important role in the diagnosis. The diagnosis is based on the combination of imaging and biopsy

[2]. The suspicion of diagnosis is important to prevent hemorrhagic complications [3].

Observation

We report the case of a 34-years-old woman, having a medical history of two seizures a year ago and which have regressed spontaneously, without any clinical or radiological exploration. The patient has been admitted to the emergency

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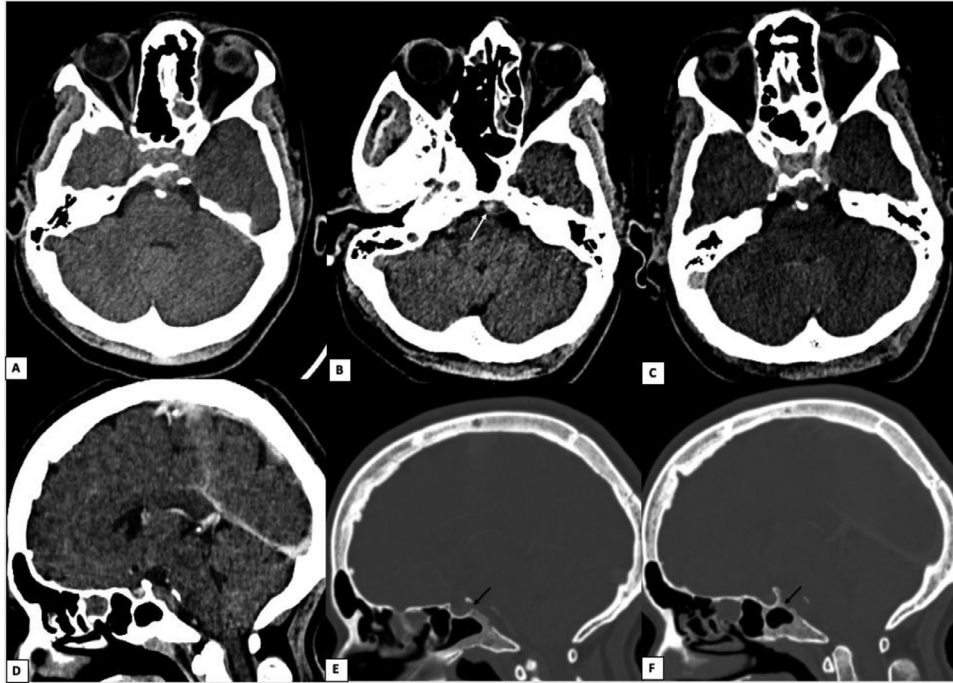


Fig. 1 - CT scan without (A, B) and with injection of the contrast (C,D) showed process of cavernous region, hyperdense and containing a phlebolith in CT scan without injection (with arrow) . It enhanced heterogeneous and moderate way in CT scan with injection and also present a bony erosion in bone windows (black arrow).

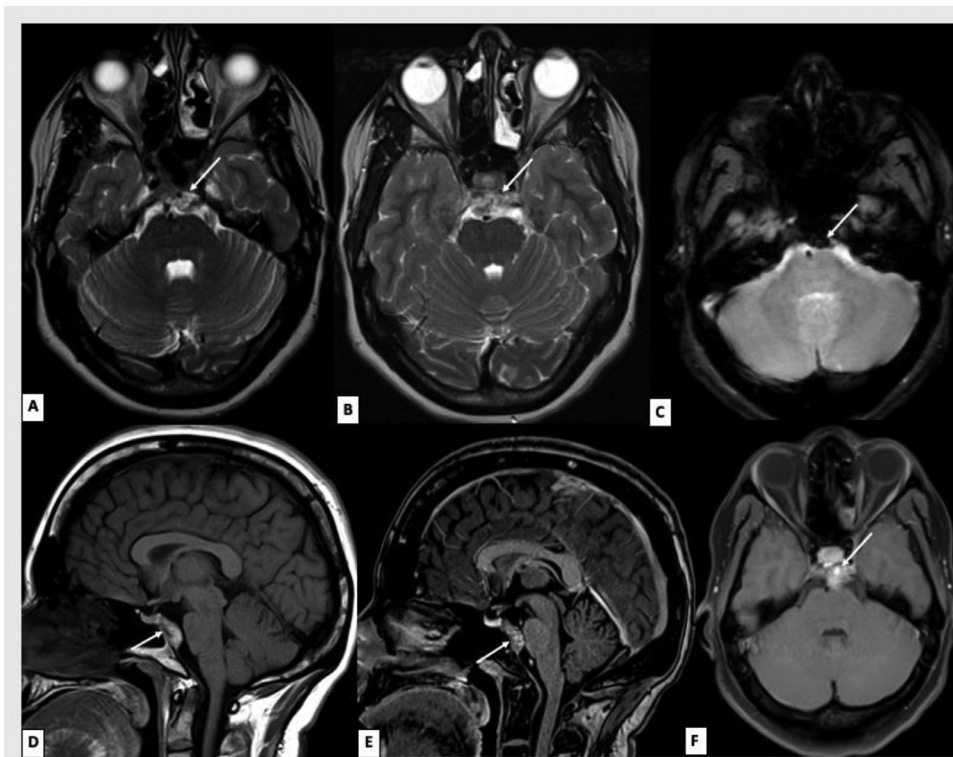


Fig. 2 - Brain MRI in T2 (A,B), T2* (C), T1 and with contrast in early time (E) and late time (F): showed process of cavernous region, in hyposignal T2, hypersignal T1 enhanced moderate and late way and containing an areas in low signal T2* (C).

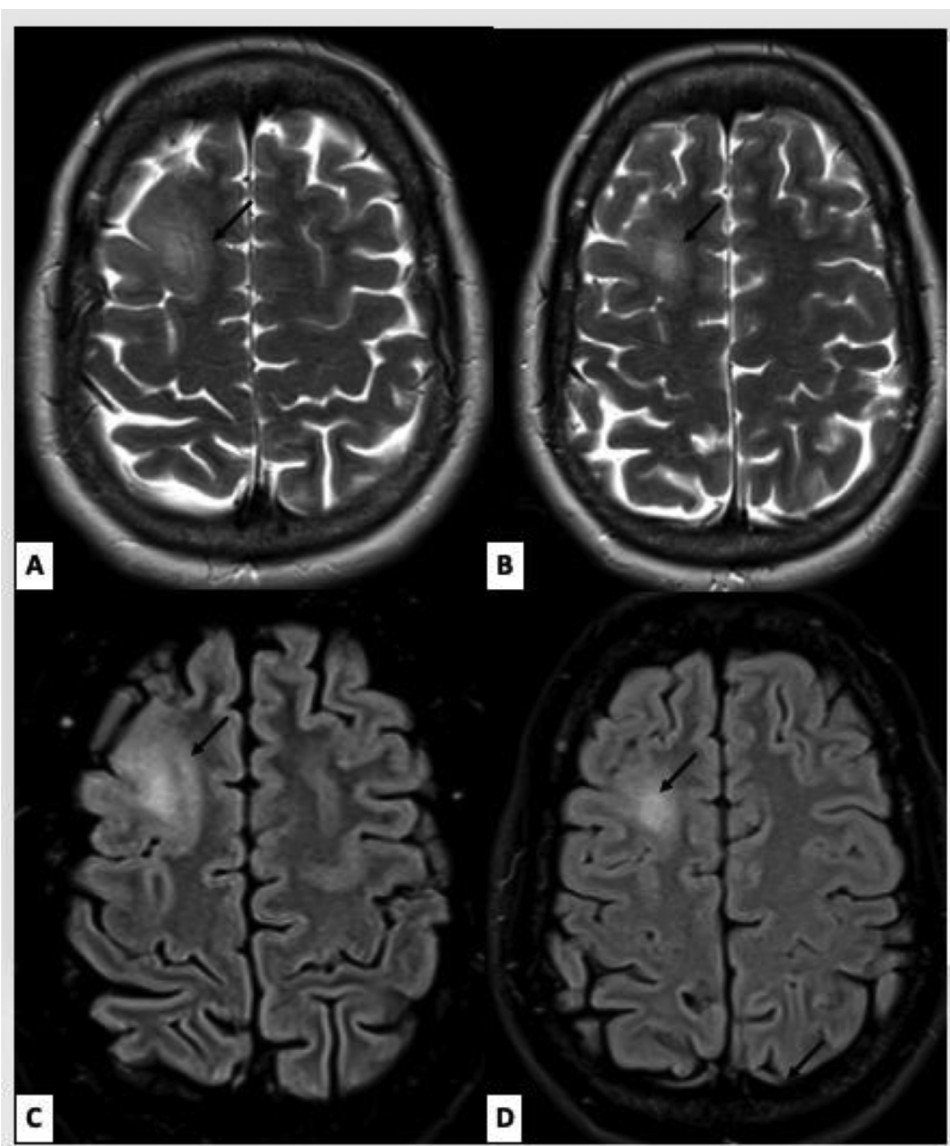


Fig. 3 – brain MRI in T2 (A, B) and FLAIR (C, D): The right frontal cortex shows a focal area of thickened cortex blurring of the grey/white matter junction and abnormal increased signal on T2 and FLAIR sequences consistent with dysplastic cortex.

for a headache with binocular diplopia for 5 days without vomiting. During the examination, the patient was conscious, afebrile, with a normal respiratory rate of 14 cycles per minute, normal heart rate (85 bpm), and with normal blood pressure measured at 135/80 mmHg. The physical examination revealed a bilateral paralysis of the fourth cranial nerve (IV). The visual acuity was normal, blood tests were all unremarkable. A CT scan without and with an injection of the contrast media has been performed and showed a process of the cavernous region, hyperdense, and containing a phlebolith in CT scan without contrast. It enhances moderately and heterogeneously in post-contrast CT scans and presents a bone erosion (Fig. 1).

Magnetic resonance imaging (MRI) revealed an enhancing mass in the left Sella turcica with extension into the posterior portion of the left cavernous sinus. The pituitary stalk

was displaced and the optic chiasm has been moved upwards. On T1-weighted MRI, the lesion was hyperintense to grey matter and enhanced heterogeneously and moderately on post-contrast T1-weighted MRI. On T2-weighted MRI, the lesion was hyperintense, with areas of low signal on T2* (Fig. 2). We have also identified a right frontal cortical dysplasia (Fig. 3). In conclusion, the diagnosis of cavernous sinus hemangioma with frontal focal cortical dysplasia was done. The surgical treatment of hemangioma was recommended, a transseptal transsphenoidal approach was used to remove the tumor and the histopathological examination was consistent with cavernous hemangioma. Diplopia is explained by invasion of the fourth cranial nerve (IV) by cavernous hemangioma because the fourth cranial nerve runs below the third cranial nerve (III) in the cavernous sinus, and through the superior orbital

fissure crossing diagonally across the levator palpebrae and superior rectus muscle to the superior oblique muscle.

Discussion

Cavernous hemangiomas are vascular malformations containing both large venous spaces with thrombosis and hemorrhage [3]. Intracranial localization is rare and representing 5%–13% of the intracranial vascular malformations and occur in approximately 0.5%–1% of the population [1,4]. They are usually present with hemorrhage, epilepsy, or rarely mass effect [5]. Extra-axial cavernous hemangioma has a different clinical presentation and can be extremely difficult to remove with high morbidity and mortality. Imaging plays a key role in the diagnosis. CT scan can show isodense or hyperdense lesion, with moderate to marked enhancement after contrast administration, with sometimes bony erosion. On MRI, cavernous sinus hemangiomas appear as well-defined masses which are hypointense or isointense in T1-WI and markedly hyperintense in T2-WI [6,7]. These lesions characteristically enhance post-contrast T1-WI [1]. The combination of bone erosion on CT marked hyper signal on T2- WI, and “salt and pepper” enhancement pattern is strongly suggestive of a cavernous hemangioma [6].

Cavernous sinus hemangiomas with sellar or intracranial extension have three patterns of para sellar growth: endophytic lateral growth, endophytic medial growth, and exophytic growth [8]. Histologically the lesion consisted of endothelial-lined vascular spaces, with only a small amount of poorly organized elastic tissue. There were also well-formed vessels consisting of feeding arteries and draining veins. Histologically, these lesions did not differ from intraparenchymal cavernous hemangiomas [6].

Differential diagnoses include adenoma, meningioma, schwannoma, and systemic neoplasm (leukemia, lymphoma). Indeed, a cavernous hemangioma may resemble pituitary adenoma, apart from the prominent hyperintensity on T2 weighted images. Regarding meningioma, it is mainly distinguished from cavernous sinus hemangioma by the presence of a broad dural base and by the dural tail. While schwannoma presents an intense and homogeneous enhancement without dural tail. On the other hand systemic neoplasm (leukemia, lymphoma) typically manifests as tissue infiltration of the cavernous sinus, hypointense on T2-weighted imaging (T2WI) with restriction of diffusion and homogeneous contrast enhancement after Gadolinium injection [9].

Cavernous hemangioma is a highly vascular lesion, surgical removal of cavernous hemangioma is often very difficult, because of the critical anatomical structures in the cavernous sinus and the risk of excessive intraoperative bleeding. If a cavernous hemangioma is suspected, the surgeon may first wish to obtain a frozen section for diagnosis and perform only a partial resection, because these lesions are dural based, and complete excision may lead to serious bleeding. Adjunctive treatment of residual cavernous hemangioma with stereotactic radiosurgery may result in excellent response and avoidance of morbidity, as has been demonstrated in recent reports [10,11].

Both vascular abnormality and cerebral cortical dysplasia are frequent causes of refractory epilepsy. Vascular abnormality associated with cerebral cortical dysplasia has been already described [12,13]. However, to our knowledge, this is the first case of cavernous hemangioma associated with focal cortical dysplasia.

Conclusion

Cavernous sinus hemangiomas are a benign vascular malformation and are extremely rare. The clinical signs of cavernous sinus hemangioma are headache, diplopia, proptosis, refractory epilepsy, or extraocular nerve palsies. The diagnosis is based on the combination of imaging and biopsy. Surgical resection is the best treatment, but it can have significant morbidity.

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