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Spontaneous Bleeding From Orbital Cavernous Hemangioma: A Case Report

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Abstract **Case Report**

Cavernous hemangioma is the most frequent primary orbital tumor seen in adults. It is a benign lesion but may compromise optic nerve function. Acute presentation with hemorrhage into the tumor is a rare occurrence with only ten cases reported in literature. We report an unusual case of a sudden painful proptosis with unilateral blindness due to acute bleeding of cavernous hemangioma, MRI revealed an intraconal soft tissue mass of the left orbit. Results: Patient underwent a transcranial approach and tumor was removed. The histopathological examination revealed a cavernous hemangioma. The patient had complete symptomatic recovery following surgery. Conclusion: Orbital hemangiomas presenting with hemorrhage is rare and should be evocated on first.

Keywords: Cavernous hemangioma, spontaneous hemorrhage, painful proptosis.

INTRODUCTION

Orbital cavernous Hemangiomas are the most common orbital tumors and known for their slow growth and benign course [1]. They are most commonly located in the intra conal compartment in the lateral aspect. Earliest clinical feature is proptosis which is slow in progression, often asymptomatic and do not warrant surgical intervention [2]. Acute bleeding of orbital cavernous hemangioma is uncommon and only 10 cases have been reported in literature. We report this rare and acute presentation of orbital cavernous hemangioma.

CASE REPORT

We report a case of a 76-years-old female with a hypertension medical history, suffered sudden onset of ophthalmic pain, redness with proptosis of the left eye, (Figure 1) and on the following day she developed a diplopia and decreased visual acuity and ptosis of the left eye as well as nausea and vomiting. These symptoms motivated our patient to visit the emergency department of our hospital when CT scan revealed an intra orbital tumor, and then our opinion was requested.

Figure 1: Clinical appearance of redness with proptosis, ptosis of the left eye and subconjunctival hemorrhage

On examination, the right eye was unremarkable, but in the left eye, there was subconjunctival hemorrhage involving the bulbar temporal and inferior conjunctiva, along with upper eyelid edema. Exophthalmometry showed a 7 mm axial proptosis of the left eye, with fundus examination revealing choroidal folds at the posterior pole and partial visual field defect in the same eye. Ocular

movement of the left eye were also limited in all directions.

CT scan revealed an extensive mass with large high-density area located in the left orbit without bone destruction (Figure 2). MRI also revealed a well-demarcated intraconal mass with partial low signal intensity within the mass as well as a low signal intensity capsule on T2 weighted imaging, and partial high signal intensity capsule on T1 weighted imaging with fluid-fluid level on both sequences, these heterogeneous signal intensities indicated acute to subacute hemorrhage within the mass. MRI with contrast medium failed to show enhancement of any major part of the mass (Figure 3 & 4).

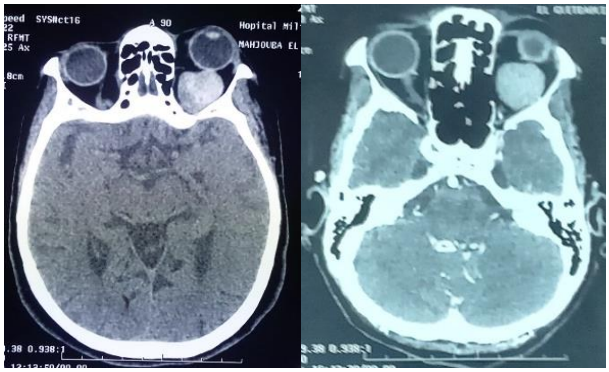

Figure 2: CT scan show an intraconal mass with large high-density area located in the left orbit

Figure 3: Axial T1-weighted (A), and T1-weighted with injection of gadolinuim (B), and fat-saturated T1-weighted with contrast meduim magnetic resonance images (C) showing the intraconal mass at different signal intensities at the hematoma and fluid-fluid levels

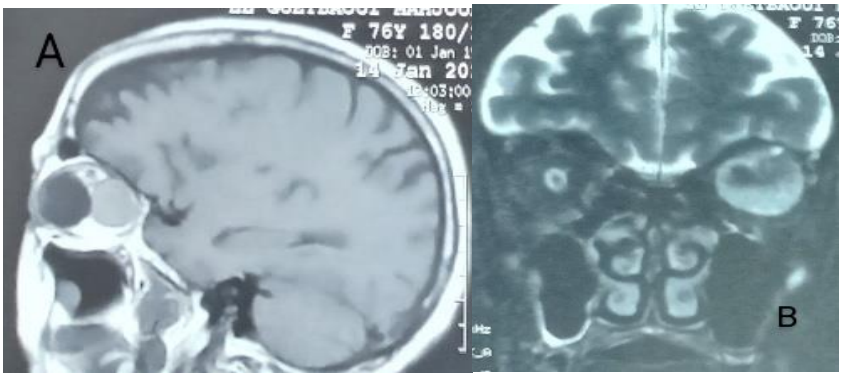

Figure 4: Sagittal T1 weighted (A), and coronal T2 weighted (B) MRI showing a well-defined intraconal mass lesion causing indentation of the posterior part of the eyeball

The patient underwent a transcranial approach with left frontal hairline scalp incision and a small frontal craniotomy was done until the sphenoid base, the roof of the orbit was cut behind the supraorbital margin to raise the orbitofrontal bone flap as a single piece. Zygomatic arch with orbital rim was then removed and lateral wall of the orbit was nibbled with KERISSON (Figure 5).

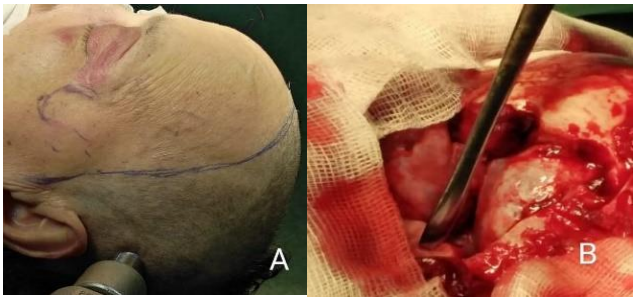

Figure 5: Position (A), and fronto-temporo-zygomatic approach (B)

The periorbita was dissected through the fat of the orbit; the medial part of the muscle cone was entered between the medial border of the superior rectus and the medial rectus muscles. The hemangioma was seen as a dark globular mass (Figure). It had a good plane of cleavage and initial dissection was done on the lateral surface, then the medial surface and then the anterior surface. The tumor was well encapsulated, had a hemorrhagic hue and was dissected away from the medial rectus muscle we were helped by cryoprobe to remove the entire mass; (Figure 6) Intra orbital and extradural hemostasis was ensured. The orbitofrontal bone flap was replaced.

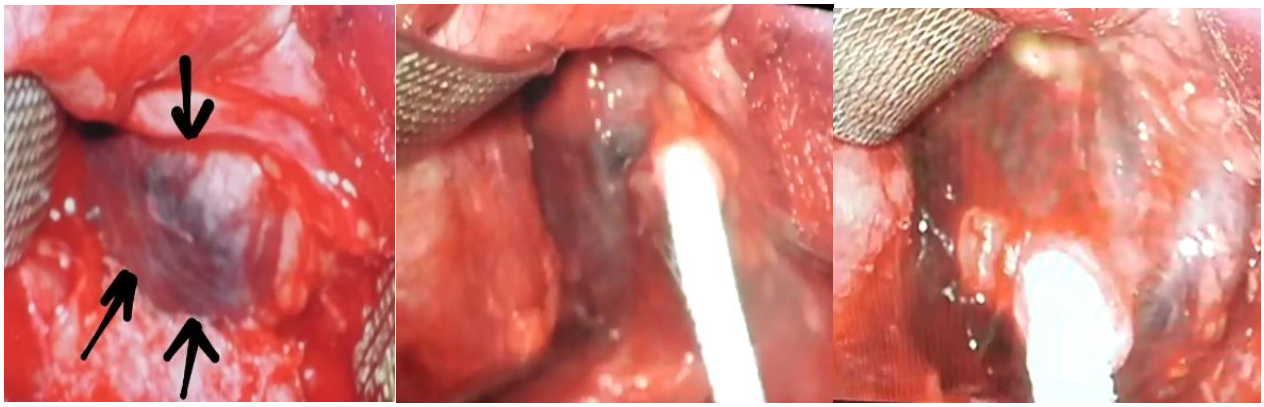

Figure 6: Cavernous hemangioma was extracted with a cryoprobe

Histopathological examination was suggestive of a tumor with congested cavernous vascular spaces lined by attenuated endothelial lining with solid areas of spindle cells with abundant eosinophilic cytoplasm. It was accompanied by a central area of organizing thrombus and chronic inflammatory infiltrate and hemosiderin deposition of the surrounding stroma- overall features being suggestive of cavernous hemangioma with hemorrhagic component. After surgery, the patient was relieved of ophthalmic pain, proptosis, and nausea; the visual field defect was also corrected. However, diplopia improved only gradually.

Figure 7: Patient seen after six months

The results of the operation 6 months after surgery showed complete (20/20) visual recovery, exophthalmos decreased, individual satisfaction, and no recurrence of the lesions.

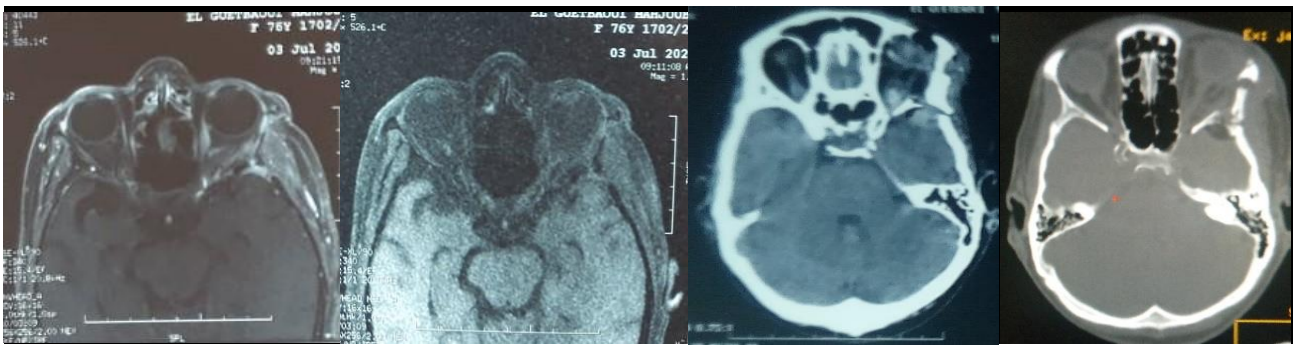

Figure 8: MRI and CT scan showing that the left orbital mass had been removed and proptosis was resolved

DISCUSSION

Cavernous hemangiomas are the most common intraorbital tumors found in adults. One large study reported an incidence of 4.3% among orbital neoplasms [3]. They are typically located in the muscle cone and the clinical course is slowly progressing without an intermittent period, and this slow growth makes the globe and orbit able to accommodate the volume expansion and developing mass, without producing disturbing symptoms [4, 5]. In the contrast to frequent hemorrhage observed with intracranial cavernous hemangiomas, spontaneous bleeding from orbital cavernous hemangioma is extremely rare. Until date, only nine cases presenting as acute hemorrhage into an orbital cavernoma have been reported [6]. Our patients here revealed rapidly onset proptosis in a few hours due to the formation of hematoma within the intraconal mass that usually do not happen in patients with orbital cavernous hemangioma.

The mechanism of spontaneous bleeding remains unclear. Some postulates include local hemodynamic changes, hypoxia [7] and bleeding from capillaries in the sinusoidal space by increased venous pressure induced by a Valsalva maneuver [1].

Subconjunctival hemorrhage and periorbital ecchymosis noted in our patient may have happened due to the blood tracking within the fascial planes of orbit, following a spontaneous orbital bleed. A reduction in visual acuity and presence of visual field defects are manifestations of a mass effect or pressure on the optic nerve and surrounding vasculature. Diplopia may occur due to extraocular muscle dysfunction.

A previous study showed that the most common symptom is exophthalmos (95%), followed by impairment of visual acuity (40%), local pain (25%), visual field defects (15%), diplopia (15%), and chronic headache (10%) [2].

Routine CT scan or MRI may reveal the orbital lesion. On a CT image, an oval or round shaped homogenous lesion with a well-defined margin may be detected and with contrast, the tumor is enhanced. However, CT alone does not allow making a definitive diagnosis. For example, Hemangiopericytoma and Neurilemmoma have similar findings to hemangioma on a CT image [8]. MRI is more sensitive and specific for the diagnosis [9].

Surgical resection of the lesion is always indicated in symptomatic patients, while asymptomatic patients should be clinically and radiologically followed up. The aim of surgical treatment is total removal of the lesion, while at the same time preserving the optic nerve and muscular structures as much as possible. [1, 4, 10] Cavernous hemangioma is a well-known lesion by ophthalmologists, which recommend anterior and lateral approach to excise the lesion. On the other hand, the neurosurgeons prefer transcranial approach, especially when the lesions involve orbital apex.

The bloc removal is possible almost in all patients, and it is necessary to avoid bleeding in the surgical field [4, 10]. The transcranial approach allow a good surgical view and exposure that are important to see the anatomical relationship in a relatively small space. The use of optic magnification will give a better precision and identification of anatomical border of cavernous hemangioma and the surgical plane [10]. Moreover, the cryoprobe, thought to be an ideal tool for hemangioma extirpation, allows for retraction of well-circumscribed tumors, and is reported to have good results in reducing intraoperative capsular rupture and bleeding [2, 11]. A bloc removal provides less complication and ensures less recurrence.

CONCLUSION

Spontaneous bleeding in an orbital cavernous hemangioma is extremely rare; However once bleeding, severe ophtalmopathy may suddenly develop, necessitating emergency surgery, as observed in the present case, and precise diagnosis of Orbital hemangioma by careful observation using MRI is essential. Generally, the surgery by transcranial approach is still proper for almost all orbital cavernous hemangiomas with good surgical outcomes.

Conflicts of Interest: The authors declare no conflicts of interest.

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