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Spontaneous regression of an intraorbital cavernous hemangioma

K. Madbouhi[1], B. Mrini[1], M. Tabchi[1], A. Amazouzi[1], N. Boutimzine[1], O. Cherkaoui[1]

[1]*Université Mohammed V Souissi, Ophtalmologie A, Hôpital des Spécialités, Centre Hospitalier Universitaire de Rabat, Rabat, Morocco.*

Abstract:
We report the case of a 60-year-old patient presenting a protrusion of the left eyeball with diplopia, which prompted the consultation. Ophthalmologic examination found retained visual acuity, left elevation deficit, painless non-pulsatile exophthalmos, eyelid edema and chemosis. Orbital CT with injection showed the presence of a cavernous hemangioma. The imaging workup is supplemented by an arteriography to specify the vascularization of the tumor with a view to surgical treatment such as lateral orbitotomy. However, the patient presented spontaneous involution, with resorption of the palpebral edema and chemosis and regression of the exophthalmos. Imaging confirmed this appearance by showing the disappearance of the initial lesion. Simple monitoring is in place. Cavernous hemangioma is a benign vascular tumor, predominantly in women, discovered incidentally or secondary to exophthalmos. The slow growth leads to a non-pulsatile, painless and reducible axial exophthalmos. In case of intra-conical localization, hyperopia, choroidal folds, papillary edema by compression of the globe can be observed. CT may be used to make the diagnosis. In case of an unusual presentation, or when the CT scan does not allow a conclusion, Doppler ultrasound and / or MRI will be useful in order to rule out differential diagnoses. The treatment consists of surgical excision in the event of clinical repercussions of the lesion. Recurrences are exceptional. Spontaneous involution is rare for orbital localization.
Keywords*:Intraorbital cavernous hemangioma; Spontaneous regression; Exophthalmos.*

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I. Introduction

Orbital cavernous hemangioma (OCH) is the most common benign vascular tumor of the orbit in adults. The pathogenicity of this vascular tumor is still debated. The diagnosis is based on a bundle of anamnestic, clinical and radiological arguments with historical confirmation. The treatment is surgical but it remains unsystematic. The authors report an atypical case of orbital cavernous hemangioma, the course of which was spontaneous regression.

II. Observation

We report the case of a 60-year-old patient, with no particular history, presenting a protrusion of the left eyeball with diplopia, which prompted the consultation. Ophthalmologic examination found visual acuity corrected to 20/20, left elevation deficit, painless non-pulsatile axial exophthalmos and normal eye tone. Examination of the appendages showed an eyelid edema and a chemosis(Figure 1a). At the slit lamp, the cornea was clear, the anterior chamber was of good depth and the iris and lens were normal. The fundus found a normal papilla. The CT with injection highlighted the presence of an intraconical oval lesion well limited with regular contours. It was strongly enhanced after injection of contrast product and measuring 10 mm by 22 mm in diameter pushing back the superior rectus muscle and respecting the optic nerve with grade III exophthalmos(Figure 2a). This CT aspect is in favor of a cavernous hemangioma. The imaging workup was supplemented by an arteriography to specify the vascularization of the tumor with a view to surgical treatment such as lateral orbitotomy. However, the patient presented spontaneous involution, with resorption of the palpebral edemaand chemosis(Figure 1b) and regression of the exophthalmos. Imaging confirmed this appearance by showing the disappearance of the initial lesion(Figure 2b). Simple monitoring was put in place.

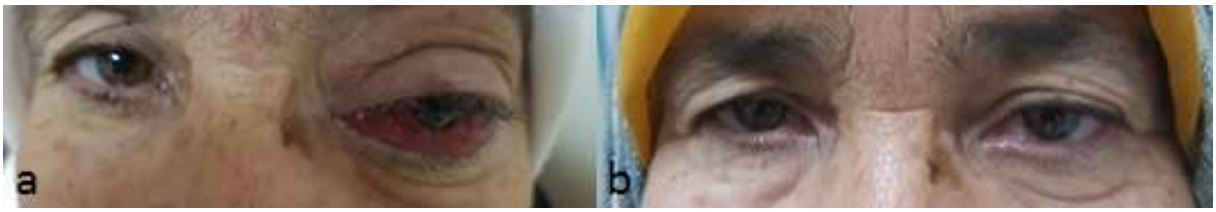

Figure 1: Image of the patient showing: a) exophthalmos of the left eye, b) complete regression of the exophthalmos a few months later.

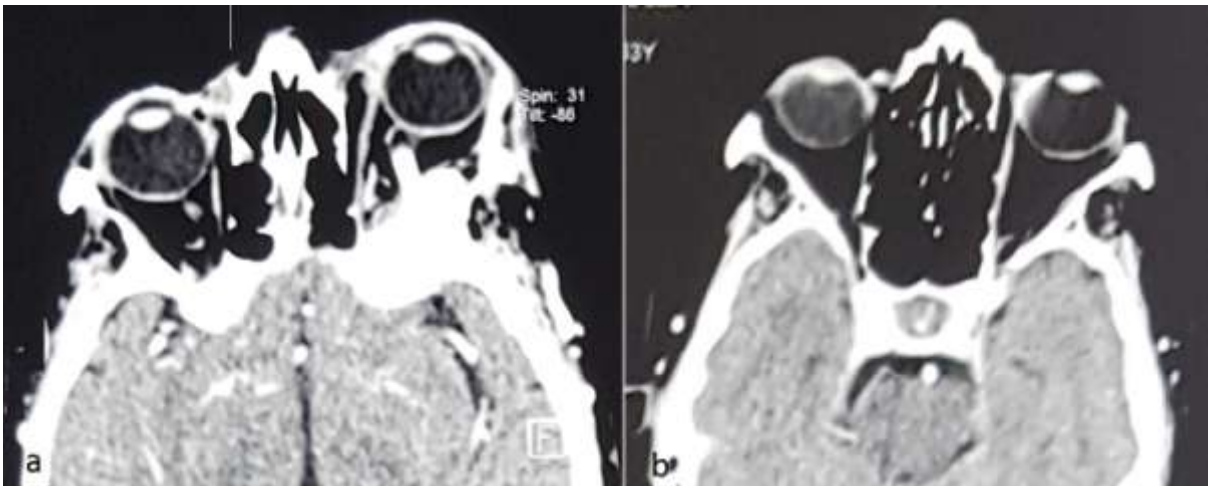

Figure 2: Orbital CT showing: a) a cavernous hemangioma of the left eye; b) complete regression of cavernous hemangioma.

III. Discussion

Cavernous hemangioma accounts for 3 to 14% of all orbital tumors according to the main published series [1] - [2], and 50 to 80% of orbital vascular tumors [3]. The origin and nature of cavernous hemangioma are still debated. OCH is classified as a vascular tumor for some, and a malformation for others [4]. It is actually a hamartoma, which haemodynamically behaves like a low-flow arteriovenous malformation, but whose anatomical features are those of a benign tumor. From a histological point of view, it corresponds to a well-circumscribed association of rather large vascular cavities, whose border is typical of those of capillaries, with rounded contours, juxtaposed in a rather fibrous and rather dense background.

Its clinical presentation is that of a slowly progressive unilateral intra-orbital mass syndrome. It represents about 25% of intraconic tumors and 7.4% of extra-conical tumors. It affects women in 70% of cases with an age of onset around 40 years [5] but is also common in children [6]. Exophthalmos is the most common warning sign, often appearing at a late stage in tumor development. It is generally progressive, painless, reducible (indicating the vascular nature of the lesion), non-pulsatile, most often axial (when the tumor is intraconic). Other clinical signs have been reported in the literature such as eyelid swelling, headache, blurring of vision, diplopia, painful motility, eye pain, enophthalmia, dizziness, visual field abnormalities, pupillary dysfunction, choroidal folds and papillary edema [7]. The topography of these hemangiomas can be variable. All intraorbital locations are possible; however, the most frequent localization is in 80% of cases intraconic, and more precisely at the level of the intraconic temporal portion of the middle third of the orbit. Bilateral and multiple localizations have been described; bilateral localization represents approximately 20 to 30% of cases [6].

If an orbital tumor is suspected, an imaging workup is essential to clarify the diagnosis. In the first line, a CT scan can confirm the presence of an intra-orbital lesion, specify certain characteristics and verify the absence of osteolysis (which would be in favor of a malignant lesion). Thus, when the questioning and the clinical examination already give rise to a suspicion of an OCH, a tomodensitometric examination can make the diagnosis by finding a rounded lesion, well circumscribed, spontaneously hyperdense, intraconic, taking the contrast in a heterogeneous way in the early stages before to homogenize. In the event of an unusual presentation, or when the CT scan does not allow a conclusion, Doppler ultrasound and / or MRI will be useful in order to rule out differential diagnoses (hemangiopericytoma, lymphangioma, schwannoma, fibrous histiocytoma, orbital varix), and will confirm in particular the low vascular flow of the lesion. An evaluation of the functional impact is also essential in order to establish a therapeutic strategy. A visual field examination should be performed routinely. Not only can alterations be detected in the absence of reduced visual acuity, but

this initial examination will serve as a reference in the event that simple monitoring is decided upon. A Hess-Lancaster test will be performed in the event of an oculomotor disorder, not systematically.

The therapeutic decision depends on the clinical impact, the location and the size of the tumor. Excisional surgery is required if the visual impairment is major. In the event of a large tumor with functional repercussions (exophthalmos, dystopia, oculomotor disorder), again surgical treatment is required. When discovered by chance with normal ophthalmologic examination, abstention with supervision is the rule. In some cases where the visual impairment is minor, the therapeutic decision will be made based on the size and location of the lesion. Spontaneous involution is rare for orbital localization; two cases have been reported in the literature [8]. Advances in imaging since the end of the 20th century have allowed important advances in the management of OCH. The scanner and MRI make it possible to specify the contours, the location, and the relationships of the tumor (optic nerve, oculomotor muscles). The precision of current investigative methods allows optimization of the choice of the first route. Surgical techniques have evolved over the past twenty years. The invasive routes by lateral or superior approach with bone flap are used less and less, to the benefit of the trans-conjunctival approach. This by micro-incision, periocular dissection to the cone, allows a precise approach to the lesion. However, it is a difficult technique with a long learning curve. The benefits for the patient are significant: reduction in operating time, scar ransom, risk of infection and postoperative pain. The lateral and superior approaches should only be offered in the event of a large tumor volume or a small tumor located at the orbital apex.

IV. Conclusion

Cavernous hemangioma is a benign tumor, but its intraconical location may warrant surgery if it compresses adjacent structures. Spontaneous involution is rare for orbital localization. The prognosis depends on the size of the lesion and its relationship to neighboring structures, but it is generally favorable.

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