

# Subconjunctival Hemorrhage as An Initial Manifestation of an Orbital Vascular Malformation

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## ABSTRACT

The present case report will describe an acute unexpected presentation of a venous lymphatic malformation of the orbit in a healthy 35-year-old female, without coagulation disorders or orbital trauma history and with a airplane travel 7 days before. The patient had an acute subconjunctival hemorrhage associated with periorbital ecchymosis, which begun 4 days before. The orbital and cranioencephalic magnetic resonance (OCMR) imaging revealed an intraconal orbital lesion inferomedial to the optic nerve with venous congestion suggestive of a venous lymphatic malformation. The proposed treatment approach was non-interventive, with clinical observation and sequential OCMR. At last observation, the patient had no relevant ophthalmologic symptoms and no significant alterations.

**Keywords:** Venous lymphatic malformation of the orbit, subconjunctival hemorrhage.

## RESUMO

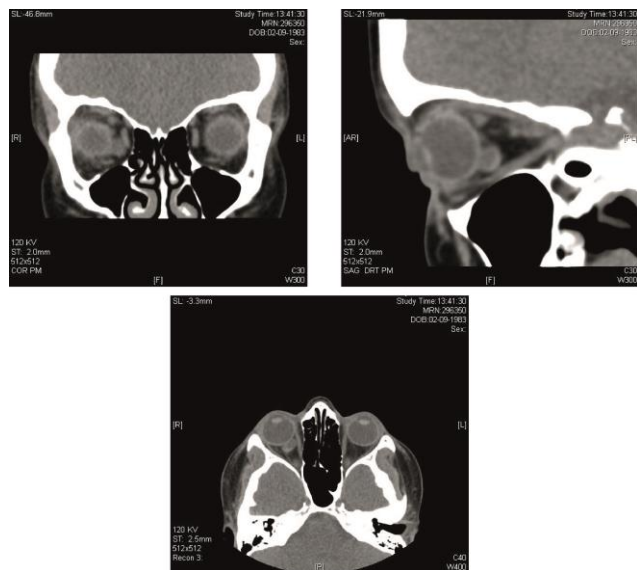
O presente caso clínico descreve uma apresentação aguda e inesperada duma malformação venolinfática da órbita. Doente do sexo feminino, saudável, com 35 anos de idade, sem doenças de coagulação conhecidas e sem história de trauma orbitário prévio, recorreu ao serviço de urgência por quadro de hemorragia subconjunctival associada a equimose periorbitária, com 4 dias de evolução. A paciente referiu ter feito uma viagem de avião 7 dias antes. A ressonância magnética cranioencefálica e das órbitas (RMC) revelou uma lesão orbitária intracónica, inferomedial ao nervo ótico e com congestão venosa, sugerindo uma malformação venolinfática. O tratamento proposto foi não-interventivo, com observação clínica e RMC seriadas. Na última observação, a paciente não tinha sintomas oftalmológicas nem alterações detetáveis no exame oftalmológico efetuado.

## CASE REPORT

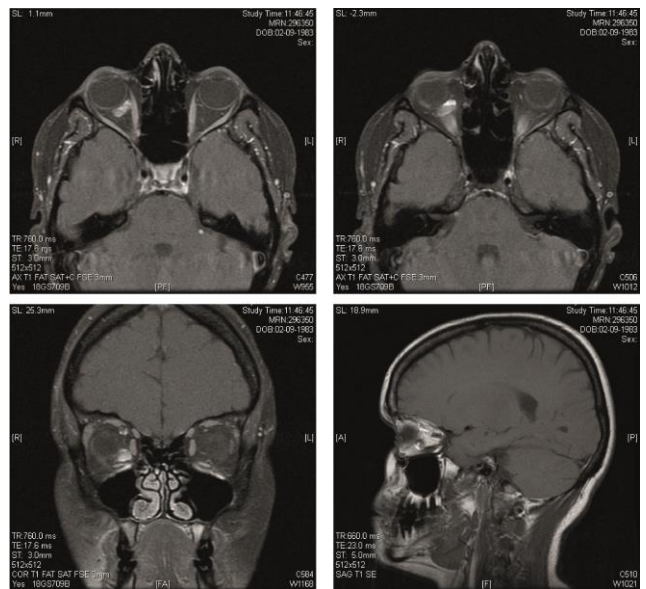
A 35-year-old female came to the emergency department with a history of right palpebral ecchymosis,

conjunctival hemorrhage of the right eye (RE) and right retroocular headache for 4 days. She made an airplane travel 7 days before and denied ocular trauma. She had no systemic or ophthalmology antecedents. Best-corrected

visual acuity of the RE was 20/20 and left eye (LE) 20/20, anterior segment biomicroscopy revealed a subconjunctival hemorrhage and inferior palpebral ecchymosis in RE, intra-ocular pressure was 15 mmHg RE and 16 mmHg LE, funduscopy had no alterations, there was no diplopia, no proptosis or palpebral retraction, ocular movements were normal. Pupilar response to light was normal. Computerized static perimetry, optical coherence tomography (OCT) and Farnsworth D-15 color test were normal. Platelet and coagulation function were normal. Computerized cranioencephalic tomography described a 9.2 mm hypodense intraconal lesion in the right orbit, inferomedial to the optic nerve, close to the inferior rectus (figure 1), which suggest a possible atypical cavernous hemangioma. Orbital and cranioencephalic magnetic resonance (OCMR) describes additional vascular engorgement of the orbit, which suggest a venous lymphatic malformation (figure 2). There were no intracerebral lesions detected. The proposed approach to this orbital finding was observation with sequential OCMR. At last observation, the patient complained of occasional right retroocular headache, had 20/20 BCVA, anterior segment biomicroscopy and funduscopy were normal.



**Figure 1** - Computerized cranioencephalic tomography described a 9.2 mm hypodense intraconal lesion in the right orbit, inferomedial to the optic nerve, close to the inferior rectus, which suggest a possible atypical cavernous hemangioma.



**Figure 2** - Orbital cranioencephalic magnetic resonance describes additional vascular engorgement of the orbit, which suggest a venous lymphatic malformation.

## DISCUSSION

The International Society for the study of vascular anomalies classification system<sup>1,2</sup> divides vascular anomalies in two basic types: vasoproliferative lesions such as hemangioma; developmental vascular abnormalities such as orbital cavernous venous malformation (named before as an orbital cavernous hemangioma<sup>3</sup>) and venous lymphatic malformations. Orbital cavernous venous malformation is the commonest benign neoplasm of the orbit in adults, which represents 5 to 9%<sup>4</sup> of the orbital lesions. It is a congenital venous malformation (the lack of ki-67 differentiates from hemangioma)<sup>5</sup> surrounded by a fibrous capsule, located more frequently within the muscle cone, lateral to the optic nerve.<sup>6</sup> There is a gender predominance, with 2/3 of the cases being detected in females.<sup>6</sup> Classic diagnosis of orbital cavernous hemangioma is made in young/middle aged adults after one gradual onset of painless proptosis over a period of years.<sup>6</sup>

In the present case, there was an acute hemorrhage manifestation and an inferomedial intraconal lesion. Due to these atypical features, cavernous venous malformation seemed unlikely.

Circumscribed choroidal hemangioma is a solitary and unilateral lesion which appears as an orange-red elevated mass in the posterior pole, with subretinal fluid, serous retinal detachment or cystoid macular edema.<sup>6,7</sup> The lack of

posterior mass in funduscopy and OCT excludes this diagnosis.

OCMR classified the lesion as a venous lymphatic malformation of the orbit.

Venous malformations of the orbit are rare, corresponding to 3 to 4% of orbital lesions in a tertiary referral center.<sup>8</sup> They are development anomalies of the vascular anlage<sup>1</sup> and can be classified in: lymphatic/no flow, venous/low flow, arterial/higher flow, venous lymphatic, arterio-venous.<sup>2,9</sup> Venous lymphatic malformations occur equally in both genders and may be noticed after the first decade of life with complaints of proptosis, globe displacement hemorrhage or deep orbital pain.<sup>8</sup> The age of our patient and the lack of proptosis are not concordant with the most common clinical picture of this diagnosis.

Half of the patients had spontaneous hemorrhage or hemorrhage due to trauma or upper respiratory tract infection.<sup>8,10</sup> In our patient, the airplane travel could be the trigger factor for the orbital bleeding, with drainage within the fascial plates of the orbit, consequent ecchymosis and subconjunctival hemorrhage.

Half of the patients with venous lymphatic malformation will need an intervention therapy because of pain, optic nerve compression, corneal exposure or cosmetic reasons.<sup>11</sup> Our patient is asymptomatic until the present moment, the proposal treatment was observation with periodic OCMR.

The possible intervention can be an embolization guided by neuroradiology, with local injection of sclerosing agents or anti-VEGF.<sup>11</sup> The surgical option can be done after or simultaneously with neuroradiology embolization.<sup>12</sup>

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None of the authors have any conflict of interests to declare.