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Spontaneous Orbital Hemmorage Revealing A Cavernous Hemangioma

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Abstract

INTRODUCTION

Orbital cavernous hemangiomas (OCH) are benign vascular tumors that commonly affect females, constituting around 4.3% of orbital tumors. These lesions often remain asymptomatic but may present with gradual proptosis or visual disturbances. Although rare, spontaneous orbital hemorrhage resulting in acute symptoms can serve as the first clinical manifestation of an OCH, necessitating urgent intervention in some cases. To date, only 11 cases of spontaneous orbital hemorrhage due to cavernous hemangioma have been reported in the literature.

Case Presentation

We present a case of a 25-year-old male who presented with acute visual loss, painful proptosis, and orbital swelling, two days following a spontaneous hemorrhage. The patient had a history of Crohn's disease, which had been untreated for two years. On examination, the right eye showed significant proptosis, impaired visual acuity, and a relative afferent pupillary defect. Orbital MRI revealed a well-defined lobulated mass with proptosis and an orbital hematoma, consistent with a cavernous hemangioma. Retinal OCT showed cystoid macular edema and chorioretinal folds. The patient was managed conservatively with NSAIDs and eye drops, leading to a progressive resolution of the hematoma over two months. Follow-up showed improvement in visual acuity and a reduction in proptosis.

Discussion

OCHs are typically diagnosed through clinical presentation and imaging, with MRI being the gold standard. Although cavernous hemangiomas are usually asymptomatic, their rupture can lead to acute orbital hemorrhage, a rare but critical complication. This case represents a rare manifestation of spontaneous orbital hemorrhage due to an OCH, which resolved with conservative management. Surgical intervention is typically reserved for cases involving significant visual compromise or progression. This case emphasizes the importance of a conservative approach in managing spontaneous hemorrhages from OCH, highlighting the potential for recovery with non-surgical treatment.

Conclusion

Orbital cavernous hemangiomas may present acutely with orbital hemorrhage, a rare complication that can often be managed conservatively. Our case illustrates a favorable outcome without the need for surgical intervention, underscoring the potential for spontaneous resolution and the importance of close monitoring in such cases. Further studies are needed to better understand the management of spontaneous hemorrhages from orbital cavernous hemangiomas.

INTRODUCTION

Cavernous hemangioma is a fairly common benign orbital venous malformation.
It is often encountered in female subjects with an estimated rate of 4.3% among orbital tumors. (1)
This vascular tumor remains often asymptomatic, it can be diagnosed following the progressive onset of an isolated proptosis or associated with other symptoms testifying to the compression of the anatomical structures of the orbit.
However, an orbital hematoma can sometimes reveal acutely the cavernous hemangioma following its bleeding, and may sometimes require a surgical approach.
This situation has only been described in the literature 13 times.

Purpose: The aim of our publication is to describe a rare acute revealing complication of cavernous hemangioma which is an orbital hematoma associated with retinal and papillary oedema , and its evolution following conservative treatment.

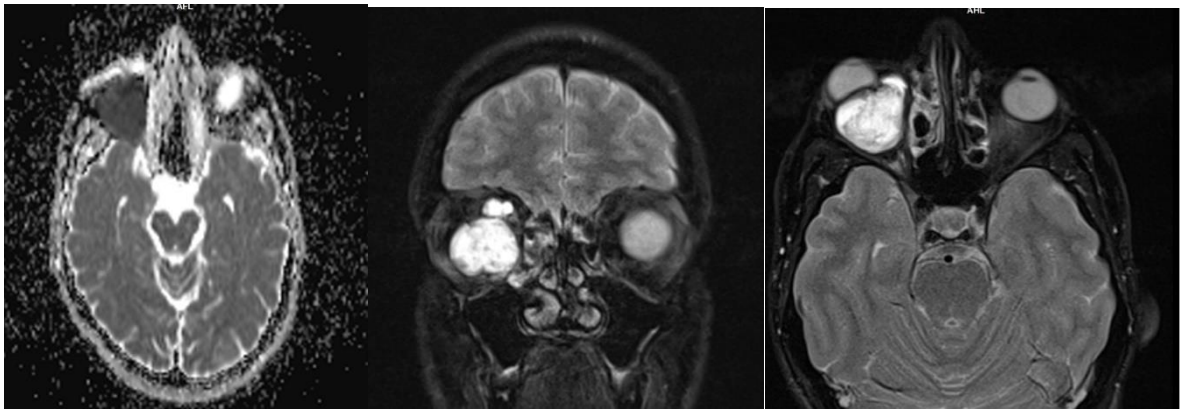
Case report:

Our case is that of a 25-year-old young man who presented to the emergency room 2 days after an acute spontaneous loss of visual acuity associated with a painful swollen red eye and severe proptosis.
He has a history of Crohn's disease followed for 7 years for which he stopped taking the treatment 2 years ago.
Inspection of the eye on admission found a large orbital bruise as well as proptosis causing a significant protrusion of the eye and scleral show.
The photo-motor reflex was lazy compared to the other eye and the pupil was in semi-mydriasis.
The patient could hardly count the fingers at 3 meters at the level of the affected eye, and had a relative limitation in eyeball mobilization.
The visual acuity of the contralateral eye was 8/10 with optical correction.
The anterior segment presented a diffuse subconjunctival hemorrhage, and the fundus revealed grade 2 papillary edema as well as oblique retinal folds of the posterior pole extended between the optic papilla and the macula.
The examination of the contralateral eye was normal.
The patient was put on non-steroidal anti-inflammatory eye drops (Diclofenac), and urgently had an orbital-cerebral MRI as well as a macular and papillary OCT.
The orbital MRI of the right eye objectified the presence of a grade 3 proptosis secondary to an orbital lesion with a mass effect on the eyeball which pushes the optic nerve up and out and comes into contact with the right muscles while respecting them.
This well-defined lobulated intra-conal tumor showed iso-intensity to the muscle in T1 and hyperintensity in the T2 sequence
Its long axes measured 36 x 30 x 28 mm and was surrounded by a thin border which was not enhanced after injection of the contrast product. *(figure 1)*
Retinal OCT showed the presence of chorio-retinal folds associated with significant cystoid macular oedema.
On the other side, the Papillary OCT confirmed the edematous infiltration of the optic nerve with a thickness of the RNFL layer exceeding considerably the normative values. *(figure2)*
The evolution was marked by the progressive resorption of the hematoma over a period of two months

with the persistence of a grade 1 proptosis and temporo-inferior dilation of the episcleral veins without strabismus nor diplopia. *(figure3)*

Visual acuity went to 6/10 under optical correction.

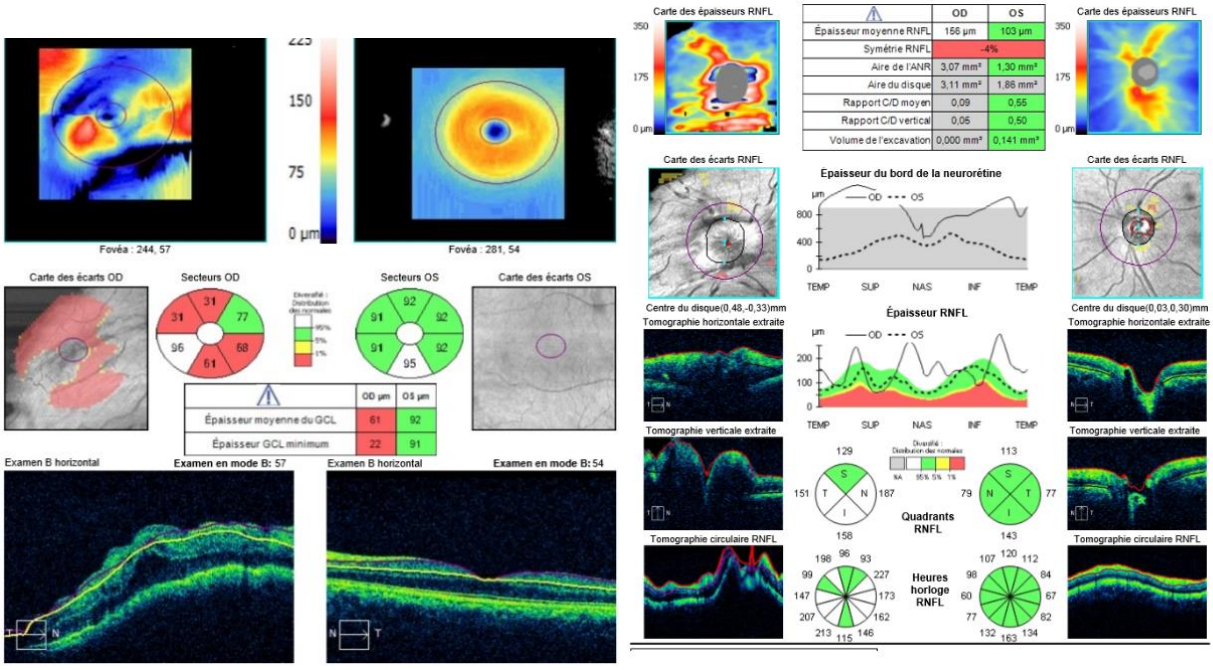
Control fundus found papillitis with well-limited papillary outline and the persistence of a few chorio-retinal folds of the posterior pole. *(figure4)*



(a) (b) (c)

Figure 1 : Magnetic Resonance Imaging of the Orbit T2+C ; (a+c) : coronal ; (b) : axial

well-defined lobulated hyperintense cavernous hemangioma showing iso-intensity to the muscle in T1 (a) and hyper-intensity in T2 (b+c) surrounded by an hypointense border constituting the hematoma (b)

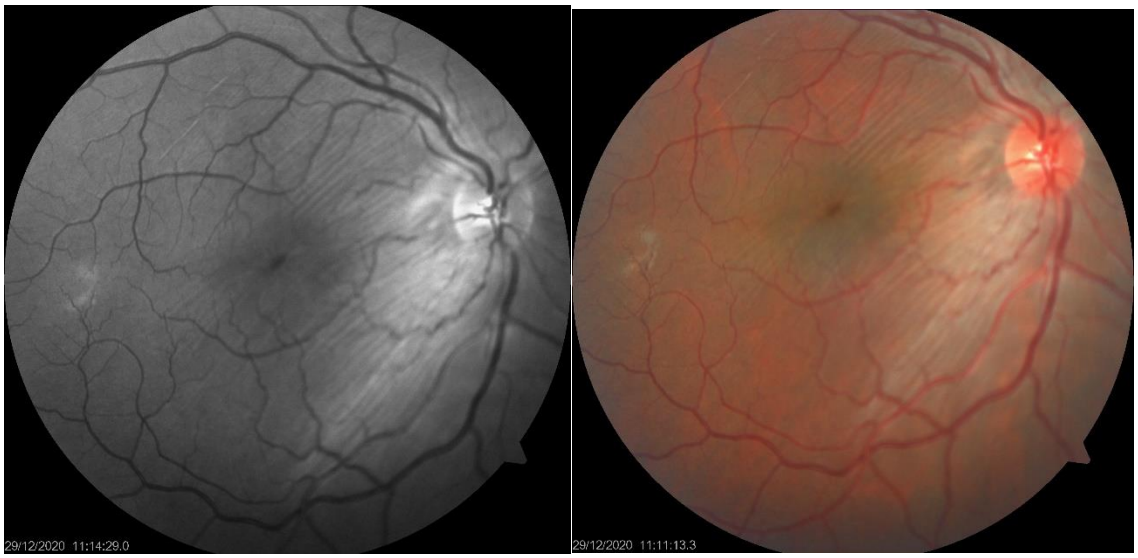


(a) (b)

Figure 2 : Macular OCT (a) showing the chorio-retinal folds and cystoid macular oedema and papillary OCT confirming the edematous infiltration of the RNFL layer.

(a) (b)

Figure 3 : Photographs of the patient taken two months later which show the persistence of proptosis of the right eye (a) as well as the dilation of the infero-temporal episcleral veins.



(a) (b)

Figure 4 : Retinophotography (a) and blue filter (b) taken two months after the acute phase, and showing the persistence of a few chorio-retinal folds as well as papillitis.

Discussion

Cavernous hemangioma is a vascular tumor in which the stroma contains cavernous spaces, the site of capillary proliferation.

It is a fairly common orbital tumor, and affects most of the times female adults in their fourth or fifth decade.

These epidemiological data were found in several studies including that of jianhua & al conducted from 1986 to 2000 and which included 214 patients with cavernous hemangioma, approximately 59% of the patients were female with a mean age of 39.4 years *(2)*

However,our patient does not belong to this population, which makes one of the particularities distinguishing of our case.

The disease may remain asymptomatic, but if it occurs two main symptoms often reveal it: the exophthalmos of insidious installation as well as the decline in visual acuity.

This was objectified in the study cited above with percentages of 76.6% and 56.1% respectively, but also in several other studies such as that of Pedro claros & al including 76 patients where 42.1% of patients presented with visual loss, 76.3% with lagophthalmos and 21.1% with exophthalmia. *(3)*

As the tumor increases in size, the clinical examination can highlight other symptoms in addition to proptosis by a simple inspection such as the deficit of ocular motility, strabismus or more rarely ptosis. The slit lamp examination of the anterior segment can also show the dilation of the episcleral veins, an exposure keratopathy ... and the fundoscopy can object other abnormalities such as choroidal folds, papilledema or even an optic atrophy.

Imagery is of great interest in the diagnosis of cavernous hemangioma, in fact orbital MRI is the gold standard and sometimes makes it possible to make the differential diagnosis with several other orbital tumors before anatomopathological confirmation. This MRI may be completed by a CT to objectify associated bone erosion.

The cavernous hemangioma is in iso-intensity to muscle in the T1 sequence, and then appears in hyperintensity after gadolinium injection.

In the event of associated bleeding as was the case for our patient, the collection signal will depend on its age and often appears as an iso-intensity or slight hypo-intensity to muscles in T1 and T2 -weighted sequences .

One of the typical aspects of this orbital tumor on MRI when its size is not very small is the heterogeneous filling at the entry of the vessels after injection of the contrast product, it follows a diffuse filling in dilated vessels.

Therefore, dynamic MRI allows us to differentiate this hematoma having radiological characters close to other tumors, and which can lead to confusion such as lymphomas, schwanomas, hemangiopericitomas, histiocytomas …

The invasive or conservative therapeutic attitude will differ depending on the type of the lesion.

(4,5)

The cavernous hemangioma has most of the time an insidious evolution which can last for years or even never clinically appear. However, in very rare cases, Its revelation can occur in various ways, with orbital bleeding leading to a retro-bulbar hematoma and mass effect as the primary cause.

In this case, an urgent surgery may be necessary in order to preserve the noble structures of the orbit namely the optic nerve and the chorio-rétina.

Since 19840 and until 2020, only 13 cases of hematoma complicating an orbital cavernous hemangioma have been reported in the literature, our patient represents the 14th one.

The table below summarizes the clinical, radiological and evolutionary characteristics of the different cases previously described in addition to our patient.

	Year of diagnosis	Age and sex	Left Eye / Right Eye	Previous symptoms	Number of days before consultation after the		Symptoms					Surgical orbitotomy	Number of days to surgery	Recovery	

					onset										
						Proptosis	diplopia	Orbital Pain	Muscle deficit	Orbital ecchymosis /subconjonctival hemorrage	Ptosis			Complete	Incomplete
Zenobii (6)	1979	M 53 years	Left eye	No	____	✓	✓	✓	✓ *(Ophtalmoplegia)*	_____	_ _ _ _	✓	18 days	✓	
Zenobii (6)	1979	M 36 years	Left eye	Yes (painless exophtalmos)	____	✓	___	✓	✓ *(slight deficit of the superior oblique muscle)*	_____	_ _ _	✓	25 days		✓ (persistance of the muscle deficit)
Zenobii (6)	1982	F 34 years	Right eye	No	2 days	✓	✓	✓	_____	✓	✓	✓	17 days	✓	
Bron (7)	2001	F 30 years	No information	No	____	✓	✓	✓	_____	_____	_ _ _	____	7 days	____	____
Jianhua (8)	2008	M 54 years	____	_____	6 days	✓	___	___	_____	_____	_ _ _	✓	6 days	✓	
Arora (9)	2011	F 40 years	Left eye	No	1 week	✓	✓	✓	✓*(limitation of upgaz*	_____	_ _ _	✓	2 weeks	____	____

									e and adduction)						
Yamamoto (10)	2012	F 61 years	Left eye	No	____ ____ __	✓	✓	✓	_____ _	_____ __	_ _ _ _	✓	9 days		✓ *(persistance of a slight diplopia improving gradually)*
Bachelet (11)	2016	F 38 years	No information	No	____ ____ _	✓	✓	✓	_____	✓	_ _ _ _	✓	2 days		✓ *(persistance of diplopia)*
Lousiraj (12)	2017	F 26 years	Left eye	No	10 days	✓	✓	✓	_____ __	✓	_ _ _ _	✓	10 days	✓	
Hegde (13)	2018	F 46 years	Left eye	No	2 weeks	✓	✓	__ __	_____ _	✓	_ _ _ _	✓	27 days	✓	
Chaoui Roqai (14)	2021	M 25 years	Right eye	No	____ ____ __	✓	✓	✓	✓	✓	No	Yes	No surgery		✓

Lee S., et al (15)	2023	F (pregnant)	Left eye	No	3 days	✓	✓	✓		✓		No	10 days	✓	
Smith J., et al (16)	2024	F 61 years	Left eye	No	5 days	✓	✓	✓		✓		No	7 days	✓	
Our case	2025	M 25 years	Right eye	No	2 days	✓	✓	✓	✓	✓	_ _ _ _	No	_____		✓

M: Male F: Female ✓ : yes _____ : No information given in the article

Cavernous sinus hemangioma surgery is not without complications; in fact several structures can be affected, namely the vessels, nerves and muscles. The three possible surgical approaches of entry are: the transconjunctival approach, the transcutaneous one and the lateral one of kronlein.

Considering the benign character and the slow evolution of this tumor, therapeutic abstention is required, and this apart from any clinical repercussions such as the alteration of the visual field, the compression of the optic nerve, the diplopia ... (17)

The orbital hemorrhage revealing the cavernous hemangioma is an acute complication responsible for a very disabling symptomatology secondary to the compression of the optic nerve and the eyeball, which requires the drainage of the blood clot and the tumor resection urgently.

After surgical excision of the cavernous hemangioma, the macroscopic examination orients the nature of this tumor. In fact , it finds a well-circumscribed tumor of purplish red color surrounded by a fibrous pseudocapsule.

Then, the histopathological examination confirms the diagnosis by revealing dilated vascular channels separated by interstitium and fibrous tissue. (17-18)

Our patient did not benefit from surgery in the acute phase despite the indication given the damage to the optic nerve and the chorio-retina. However, the evolution a few months later with the topical treatment with non-steroidal anti-inflammatory eye drops, the patient improved his visual acuity, the proptosis regressed considerably, the chorio-retinal folds decreased in size and depth, and only a slight edematous infiltration of the optic nerve persisted, which constitutes a phenomenal evolution .

Apart from the surgical resection of the tumor and therefore the treatment of the causative agent responsible for the bleeding, our case is the ultimate proof that acute bleeding of low abundance although exerting a mass effect can resolve itself and quickly without indelible repercussions on the noble structures of the eye.

Conclusion

Cavernous sinus hemangioma is an orbital lesion frequently observed in adults, its progressive evolution makes it a benign lesion often respected, except in cases where the clinical repercussions oblige practitioners to treat it surgically.

The bleeding of this lesion is very rare, and has only been reported in the literature 13 times.
Orbitotomy for drainage of the hemorrhage and tumor resection was performed in 10/11? cases out of 11 with the exception of our case where we only carried out simple clinical monitoring under medical treatment while waiting for the spontaneous absorption of the hematoma.
In conclusion, the description of our case shows one of the rarest clinical pictures revealing cavernous hemangioma, but also the indication of a conservative treatment with rigorous monitoring in the event of minimal bleeding while waiting to schedule the surgery.

References

1. Henderson GW. Vascular hamartomas, hyperplasias, and neo-plasms. In Henderson GW, editor. Orbital tumors. New York:Raven Press; 1994;94–100
2. Jianhua Yan [1], Zhongyao Wu ,Cavernous hemangioma of the orbit: analysis of 214 cases
3. Pedro Clarós , Emmanuel Choffor-Nchinda , Marta Lopez-Fortuny, Andres Claros & Salvador QuintanaOrbital cavernous haemangioma; profile and outcome of 76 patients managed surgically, Published online: 01 Jun 2019
4. Ajay Hegde [1], G Lakshmi Prasad , Girish Menon , Padmapriya Jaiprakash
Spontaneous orbital haemorrhage secondary to cavernous haemangioma - a case summary and review of literature
5. A Diamantopoulou, Ch Damianidis, V Kyriakou, N Kotziamani, M Emmanouilidou, F Goutsaridou, I Tsitouridis
6. Orbital cavernous hemangiomas: ultrasound and magnetic resonance imaging evaluation
7. Zenobii M, Galzio RJ, Lucantoni D, Caffagni E, Magliani V. Spontaneous intraorbital hemorrhage caused by cavernous angioma of the orbit. J Neurosurg Sci. 1984;28(1):37–40.
8. C. Born, J. Rademaker, N. Hosten & R. Felix (2001) Hemorrhagic cavernomaor ruptured dermoid of the orbit: Diagnosis with MRI, Orbit, 20:4, 291-295, DOI: 10.1076/orbi.20.4.291.2616
9. Yan J, Wang X. Cavernous hemangioma with rapidly developing proptosis. IntOphthalmol 2008;28(2):125–6.https://doi.org/10.1007/s10792-007-9115-0.
10. Arora V, Prat MC, Kazim M. Acute presentation of cavernous hemangioma oftheorbit.Orbit2011;30(4):195–7.https://doi.org/10.3109/01676830.2011.579684.
11. Yamamoto J, Takahashi M, Nakano Y, et al. Spontaneous hemorrhage fromorbital cavernous hemangioma resulting in sudden onset of ophthalmopathyin an adult–case report. Neurol Med Chir(Tokyo) 2012;52(10):741–4.
12. Bachelet J-T, Berhouma M, Shipkov H, Kodjikian L, Jouanneau E, Gleizal A.Orbital Cavernous Hemangioma Causing Spontaneous CompressiveHemorrhage. J Craniofac Surg 2018;29(3):706–8.https://doi.org/10.1097/SCS.0000000000004285.
13. Louisraj S, Ponnudurai T, Rodriguez D, Thomas PA, Nelson Jesudasan CA.Cavernous hemangioma of the orbit: an unusual acute presentation. Int MedCase Rep J 2017;10:255–9.https://doi.org/10.2147/IMCRJ.S133284.
14. Ajay Hegde, G. Lakshmi Prasada,⇑, Girish Menona, Padmapriya Jaiprakash
15. Spontaneous orbital haemorrhage secondary to cavernous haemangioma– a case summary and review of literature
16. Y. Chaoui Roqai, M. Aachak, Y. Malek, A. El Khoyaali, Y. Mouzari, F. El Asri, K. Reda, A. Oubaaz
17. Bleeding from an orbital cavernous hemangioma: A rare presentation Doi : 10.1016/j.jfo.2020.04.045

18. Lee S., et al., "Orbital Cavernous Hemangioma in a Pregnant Woman: A Rare Presentation and Management," Journal of Clinical Medicine, 2023; 43(5): 78-85.
19. Smith J., et al., "Spontaneous Orbital Hemorrhage from a Cavernous Hemangioma: A Case Report," Journal of Clinical Medicine, 2024; 45(7): 123-130.
20. Ansari SA, Mafee MF: Orbital cavernous hemangioma: role of imaging. Neuroimaging Clin N Am 15:137-158, 2005.3
21. Pol J Radiol 73(3):59-62, 2008 Smoker WRK, Gentry LR, Yee NK, Reede DL, Nerad JA: Vascular lesions of the orbit: more than meets the eye.