

Eyelid Swelling as a Typical Manifestation of Ocular Sarcoidosis

A. Oualime², T. Mahloute², F. Slimani^{1,2}

¹Faculty of Medicine and pharmacy, Hassan II University of Casablanca, P.O. box5696, Casablanca, Morocco

²Departement of oral and Maxillo-facial surgery, 20 august hospital, Ibn Rochd University Hospital, P.O. box2698, Casablanca, Morocco

Abstract: *We report a case of a 56-year-old woman with no significant pathological history, who had a right upper palpebral tumefaction gradually increasing in volume for 5 months. Clinical and paraclinical assessments led to a palpebral tumor without nature precising.. The diagnosis of sarcoidosis without systemic involvement was confirmed by the anatomopathological examination of the biopsy excision of the mass. The palpebral involvement without systemic involvement is an exceptional presentation, only a few cases have been reported in the literature.*

Keywords: Eyelid; sarcoidosis; orbit

1. Introduction

Sarcoidosis, or Besnier-Boeck-Schaumann's disease, is a multi-systemic granulomatous inflammatory disease of unknown etiology, most often revealed by pulmonary signs. Eye involvement is sometimes indicative of the disease [3].

Anterior uveitis is the most common manifestation of ocular sarcoidosis and can be affected by cornea, optic nerve or eyelid [2], [10].

The palpebral involvement without systemic involvement is an exceptional presentation, only a few cases have been reported in the literature [7], [10].

We report the case of an isolated palpebral involvement initially presented as a palpebral tumor.

2. Case Report

A 56-year-old woman presented with no significant pathological history, who had a right upper palpebral tumefaction gradually increasing in volume for 5 months, associated with palpebral ptosis.

Clinical examination resulted in a right upper palpebral mass of firm non-painful consistency at palpation measuring 1.5 cm, non-inflammatory, movable with respect to both planes with conjunctival hyperaemia of the right eye [Figure 1], [Figure 2], Without impairment of ocular motility or visual acuity.



Figure 1: Right palpebral swelling and right upper palpebral ptosis



Figure 2: Right palpebral swelling and right upper palpebral ptosis

An ocular ultrasound which showed an upper ocular formation at the right upper palpebral level, a rather limited hypoechogenic formation with a hyperechogenic center with peripheral vascularization measuring 11/6 mm. A CTScan showed an infiltrating tissue process in the upper right

eyelid enhancing the contrast product [Figure 3]. A frontal chest X-ray was normal with a standard biological check-up with no special features.

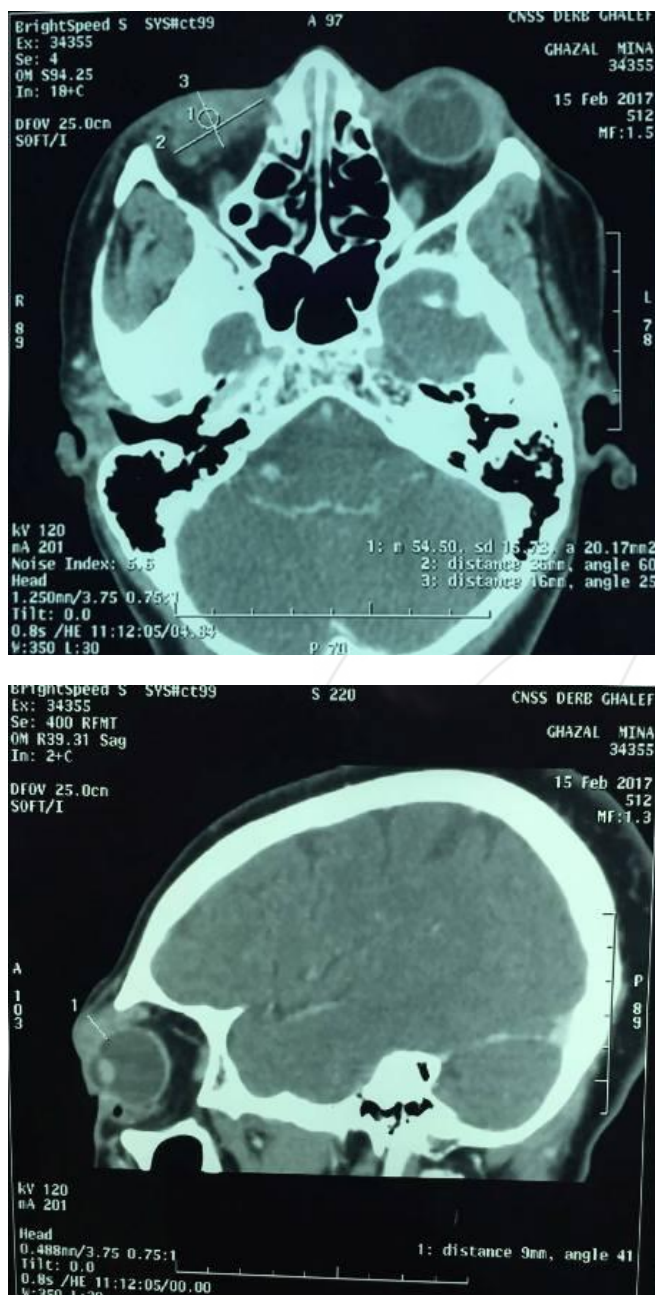


Figure 3: Craniofacial CT: A: axial cut, B: sagittal cut

The surgical exploration under general anesthesia, by medio-palpebral upper right has revealed a yellowish oval process adhering to the levator muscle of the upper right eyelid. The patient underwent a biopsy excision of the mass. Immediate postoperative follow-up was simple except for a slight swelling and upper right palpebral ecchymosis without ptosis [Figure 4].



Figure 4: One week after tumor excision

The anatomopathological examination of the surgical specimen had been inflammatory nodule, that is the site of a non-necrotizing tuberculoid granulomatous inflammation suggestive of sarcoidosis. The patient was addressed in internal medicine or a systemic assessment was carried out without the discovery of another localization. The patient was treated with degressive corticosteroids with regular follow-up in consultation.

3. Discussion

Sarcoidosis is a multi-systemic granulomatosis of unknown cause affecting preferentially young women, the preferred locations are pulmonary and mediastinal [5].

Although cutaneous sarcoidosis accounts for 12 to 27% [1], [8]. Palpebral localization without other systemic signs is rare [4], [7].

The most common manifestations are small papules, although large nodules, ulcerated nodule [7].

Yanardag and Pamuk found 13% eye disease in a series of 516 patients with sarcoidosis, but palpebral involvement was not observed [9].

A retrospective study by Dios Castro et al in 18 patients with ocular sarcoidosis showed no palpebral involvement [13].

Although in the literature there are rare cases of palpebral involvement of sarcoidosis, Souza Filho described a palpebral involvement of sarcoidosis without systemic signs in a 73-year-old woman [7], Flach described the same [11].

Biswas et al reported a case of orbital sarcoidosis with eyelid Swelling and diplopia [7].

The palpebral involvement as the initial manifestation of sarcoidosis was reported by Rosa and Unsold [12].

The others reported a combination of bilateral orbital involvement with upper palpebral mass.

Our 56-year-old patient with no previous pathological history who had palpebral swelling and the biopsy was in favor of sarcoidosis without other systemic manifestations.

Yet the microscopic aspect of inflammatory granulomatosis is not pathognomonic of sarcoidosis [4], other pathology may have the same microscopic appearance as tuberculosis, leprosy. These pathologies can be the differential diagnosis[5].

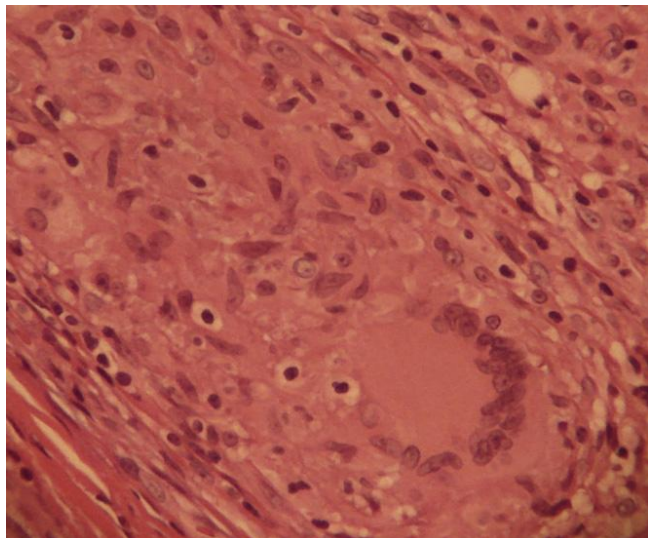


Figure 4: Histological section, staining of hematoxylin-eosin $\times 380$: granuloma giganteo-celular without caseous necrosis[5].

In our patient, the Angiotensin I-converting enzyme (ACE) rate was at the limit of normal, this result can be explained by the fact that the patient was under corticosteroid therapy and corticosteroid therapy was shown to decrease the ACE level [6]. In some cases, palpebral involvement as an initial manifestation of sarcoidosis has been observed for years before other organs have been reached, that why all patients must be systematically surveyed with clinical monitoring and paraclinical examination to diagnose a secondary localization [7].

The case of Souza Filho was monitored for 3 years during which the patient did not present other systemic sites [7]. Our present patient is followed in internal medicine department and so far she has not developed another systemic manifestation knowing that her palpebral mass has disappeared and she is still under depressive dose corticosteroids.

4. Conclusion

The diagnostic approach to eyelid swelling should include the systematic search for sarcoidosis because of the associated therapeutic involvement, although this etiology remains exceptional.

References

- [1] Descamps, F. Bouscarat : Manifestations cutanées de la sarcoïdose. *Annales de dermatologie et de vénéréologie* (2016) 143, 39-50
- [2] P, Robert . M, Baughman, : Ocular Sarcoidosis. *Semin Respir Crit CareMed* 2010;31:452–462.
- [3] C. Brunellea, S. Ballya, J. Provencal ,C. Lecomte : Pseudotumeur inflammatoire orbitaire révélant une sarcoïdose. *J f d’oph*(2016) 39, e179-e181
- [4] A. Atig, A. Guiga , A. Bouker ,N. Ghannouchi , F. Bahri: Atypical sarcoidosis. *Revue de Pneumologie clinique* (2017)
- [5] D. Nguyen Van Nuoï, Q. Zhang, E. Finke, R. Hage, T. David : Tuméfaction unilatérale de la glande lacrymale révélatrice de sarcoïdose à 85 ans. *J F d’ophthalmol*(2010) 33, 342.e1—342.
- [6] O. Badarani : Anti infl ammatory treatments and other treatments in sarcoidosis. *Revue des Maladies Respiratoires Actualités* (2016) 8, 156-158
- [7] O. Badarani : Anti infl ammatory treatments and other treatments in sarcoidosis. *Revue des Maladies Respiratoires Actualités* (2016) 8, 156-158
- [8] Cacciatori M, McLaren M, Kearns PP.: Sarcoidosis presenting as a cutaneous eyelid mass. *Br J Ophthalmol*.1997;81:329–330.
- [9] H , Yanardag, O, Pamuk . Lacrimal gland involvement in sarcoidosis. *Swiss Med Wkly*. 2003;133:388–391.
- [10] S. Pasadhika, T. Rosenbaum : Ocular Sarcoidosis. *Clin Chest Med* 36 (2015) 669–683
- [11] Flach A. Symblepharon in sarcoidosis. *Am J Ophthalmol*. 1978;85:210–214.
- [12] Rossa V, Unsoïld R. Initial manifestation of sarcoidosis as a palpebral or para-bulbar tumor. [English translation] *Fortschr Ophthalmol*. 1989;86:162–163.
- [13] E .Dios Castro: Sarcoidosis ocular. Retrospective study of 18 cases. *Arch Soc Esp Oftalmol*. 2002;77(6):301–308.