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Case Report

Chronic unilateral anterior scleritis, think about a herpetic origin: A case report

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ABSTRACT

Scleritis is a very heterogeneous group of diseases responsible for ocular inflammation of varying severity, the evolution and prognosis of which depend on the etiology but also on the appropriate treatment.

We report the case of a 15-year-old female patient, without any notable general history, followed in consultation for 3 months for a nodular scleritis not improving under usual treatments. She was reconsulted when her symptoms worsened and the examination revealed a minimal reaction of the anterior chamber, in addition to the nodular scleritis localized in the temporal region, with the notion of a vesicular eruption in the perioral region on the same side of the ocular involvement and preceding the ocular symptoms by one week, consistent with a cutaneous herpes.

The patient was put on oral Aciclovir (800 mg, 3 times a day) with a clear improvement marked by a decrease in pain and redness, which improved again after the introduction of oral corticoids. The existence of an extra-ocular sign of herpes and the good response to antiviral treatment, thus confirming the herpetic etiology of this chronic nodular scleritis.

Scleritis is caused by herpes in about 5% of cases. The clinical picture is either ophthalmic herpes zoster with associated scleritis (VZV), or diffuse anterior scleritis, unilateral in 80% of cases, related to herpes simplex. The diagnosis can be confirmed by local swabs, or more often by an antiviral therapeutic test.

1. Introduction

Scleritis is a rare ocular inflammatory disease caused by occlusive vasculitis of the deep episcleral plexus with a risk of ischemia and necrosis [1]. The inflammation is deep, painful, potentially chronic and sometimes blinding.

Their etiologies are dominated by autoimmune involvement. Although rare, an infectious etiology should be considered in any patient with scleritis, especially those with long-standing chronic inflammation or failure to respond to standard therapies or postoperative necrotizing scleritis. Infectious scleritis accounts for approximately 8% of the etiologies and is mainly of herpetic origin (varicella-zoster virus and herpes virus) [2]. When the diagnosis of herpetic scleritis is suspected but cannot be confirmed bacteriologically, a trial antiviral treatment can be considered, given the excellent response in proven herpetic scleritis [3,4], with therapeutic test value.

We report the case of a patient followed for 3 months for a chronic scleritis resistant to the usual treatments, for which herpetic etiology was the cause.

2. Clinical observation

Our work consists of a single case report and has been reported in accordance with SCARE 2020 criteria [5].

The patient was 15 years old, with no notable general history. She has been wearing optical correction for 3 years, regularly followed. She was also followed up in consultation for 3 months for the notion of repeated right red eyes, diagnosed as nodular anterior scleritis of an as yet unlabelled etiology despite a complete immunological work-up, which did not improve under usual treatment. She was readmitted by her internist for intensification of clinical signs with the notion of a decrease in visual acuity for 48 hours and slight ocular pain in

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abduction.

On admission, she had a corrected visual acuity of 9/10 bilaterally and an ocular tone of 14 mmHg on the air tonometer. Slit lamp examination of the surface of the right eye revealed conjunctivo-scleral hyperemia localized in the temporal region with the presence of a nodule of approximately 4 mm, non-mobilizable, with no focus of necrosis or scleromalacia (Fig. 1). The cornea was clear without fluorescence and a fine cellular tyndall was present in the anterior chamber. The rest of the examination was without abnormality, in particular the fundus did not show any retinal or choroidal folds or papillitis. The examination of the left eye was normal.

The diagnosis of chronic nodular anterior scleritis was retained in view of the examination data and a negative neosynephrine test. The examination revealed a vesicular eruption in the perioral area on the same side of the eye, which preceded the eye symptoms by one week and was consistent with cutaneous herpes.

The general examination did not reveal any other cutaneous-mucosal localization of herpes. The patient was put on ACICLOVIR per os at a dose of 800 mg 3 times/day with local hygienic measures for her perioral herpes.

The evolution was marked by a clear improvement with a decrease in pain and redness after 24 hours. The addition of oral corticosteroid therapy 48 hours after the start of the antiviral treatment improved all clinical signs with regression of the nodule after 2 weeks (Fig. 2). A dose reduction of Aciclovir was then started. However, the patient stopped her treatment on her own without progressive degression or maintenance dose; she reappeared in emergency one week later with the onset of contralateral involvement (left eye) in the form of acute temporal scleritis, without any individualizable nodule (Fig. 3). This necessitated resumption of treatment at the loading dose for 1 month with 2 weeks of oral corticosteroid therapy. There was no relapse during degression for 3 additional months and 6 months of maintenance dose at 800 mg daily. The course was marked by complete recovery bilaterally after a total of 4 months of background therapy (Fig. 4).

In sum, the etiology of this chronic nodular scleritis in our patient is herpetic. After one year of follow-up, there were no complications or relapses.

3. Discussion

Scleritis is a rare ocular inflammatory disease caused by occlusive vasculitis of the deep episcleral plexus with a risk of ischemia and necrosis [1]. Scleritis must be differentiated from episcleritis, which is an isolated, mild to moderate inflammation of the superficial tissues, rarely associated with systemic disease. Scleritis is a very heterogeneous group responsible for the inflammation. This is explained by the variability of the site of the attack (anterior or posterior), of the clinical presentation (diffuse, nodular or necrotizing), of the etiology which can be autoimmune (most frequent cause), infectious (viral or bacterial) or iatrogenic and finally of the evolution (under targeted or probabilistic adapted treatment).

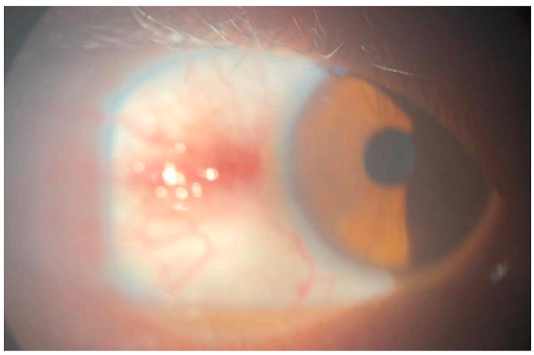

Fig. 1. Appearance of nodular scleritis localized in the temporal region.

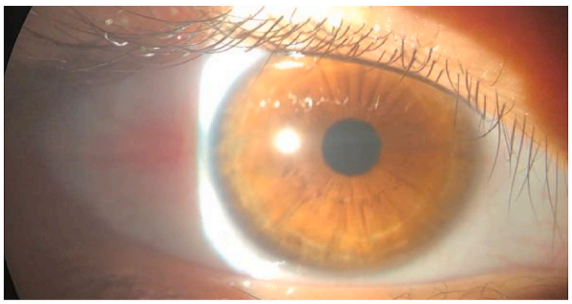

Fig. 2. Appearance after 2 weeks of treatment: good evolution and regression of inflammation.

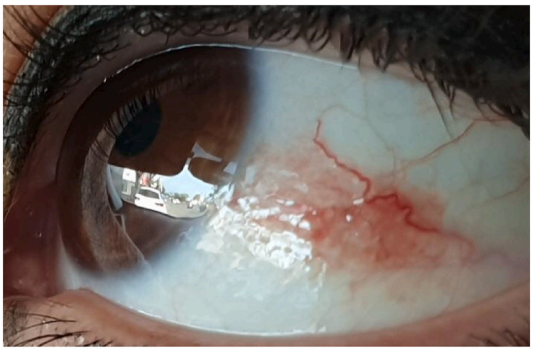

Fig. 3. Appearance of temporal scleritis of the left eye.

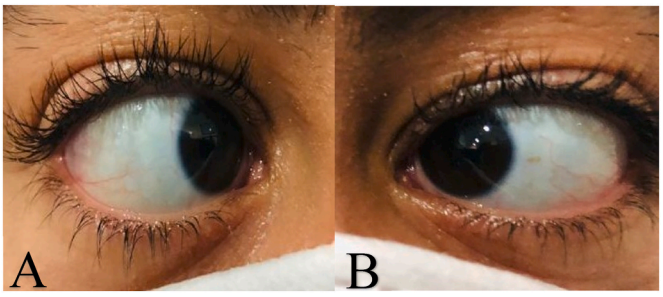

Fig. 4. Appearance after 4 months of background treatment (A: RE and B: LE).

Scleritis is associated with general pathologies in 40–50% of cases [6,7]. The etiologies are dominated by systemic diseases (rheumatoid arthritis and necrotizing vasculitis [30–40%]) and infections (mainly herpes virus [10%]). Scleritis is caused by herpes in about 5% of cases. The clinical picture is either ophthalmic herpes zoster with associated scleritis (VZV) or diffuse anterior scleritis, unilateral in 80% of cases, related to herpes simplex [2]. The unilateral character and the anterior nodular type as well as the association with interstitial keratitis or uveitis are in favor of an infectious origin while the bilateral character and the female sex seem to predict a systemic origin [1].

In our case, the patient presented with a unilateral nodular anterior scleritis, with minimal anterior chamber reaction, which was in favor of an infectious involvement, especially in view of the non-improvement under the usual anti-inflammatory treatments.

Herpetic etiology is the most frequent viral cause, accounting for about 8% of all etiologies; however, it remains unrecognized for a long time, which leads to a very careful examination in search of signs in favor of herpes. Also, it is important to perform a scleral biopsy in the case of any scleritis that is resistant to the usual treatment, with anatomopathological examination and the search for the viral genome using

the PCR technique [3]. Moreover, the existence of a suggestive sign dispenses with serology, which remains positive in most cases.

In our case, perioral cutaneous herpes was a very favorable sign. We did not resort to scleral biopsy or serology because of the presence of suggestive signs but also and above all because of the improvement under antiviral treatment, which brings diagnostic certainty.

The treatment of herpetic scleritis is based on systemic antiviral drugs. The most commonly used are ACICLOVIR and VALACICLOVIR (which remains a prodrug of Aciclovir) [2]. Thus, if biopsy is not possible and the suspicion of herpetic infection is strong (i.e., unilateral, not responding to steroids), a trial of ACICLOVIR (800 mg 3 times daily orally) is suggested and consideration of herpes virus antibodies should be given. If there is evidence of herpes zoster virus infection (e.g., herpes zoster ophthalmicus), it is acceptable to use ACICLOVIR 800 mg 5 times daily or FAMCICLOVIR instead. However, the dose of Aciclovir varies (800 mg 3 to 5 times daily). This dosage corresponds to 1–2 g/day of Valaciclovir. Gonzales et all [4] report a regression of inflammation in less than 3 weeks in simple forms (3–8 weeks in more chronic forms). After 1 month without signs of inflammation, the dosage is gradually decreased to the maintenance dose to avoid inflammatory recurrence.

The risk of recurrence of herpetic scleritis is often high, requiring the maintenance of antiviral coverage.

4. Conclusion

Scleritis is an ocular pathology that is always a cause for concern. It constitutes a heterogeneous group of inflammation of the sclera, requiring a rigorous multidisciplinary management (ophthalmologist, internist, immuno-biologist), because of the frequency of the autoimmune etiology, this, should not in any case make us forget the infectious causes essentially herpetic especially in case of ocular herpetic history or other. The diagnosis can be confirmed by local swabs or, more often, by an antiviral therapeutic test.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

Issiaka Moctar: Corresponding author, writing the paper. Abounaceur Amina: writing the paper. Aitlhaj Jihane: writing the paper. Mchachi Adil: writing the paper. Benhmidoune Leila: writing the paper. Rachid Rayad: writing the paper. El Belhadji Mohamed: Correction of the paper.

Registration of research studies

1. Name of the registry: research registry
2. Unique Identifying number or registration ID: 6910
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

ISSIAKA MOCTAR.

Declaration of competing interest

Authors of this article have no conflict or competing interests. All of the authors approved the final version of the manuscript.

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