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Vernal Keratoconjunctivitis with Giant Papillae as Manifestation of Post Streptococcal Syndrome: Report of the First Case and Review of the Literature

Said Iferkhass*, Nihad El halouat, Hatim Boui, Anas Bouassel, Abdalkader Laktaoui
Ophthalmology Service Military Hospital My Ismail Meknes Morocco

Case Report

***Corresponding author**
Said Iferkhass

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Abstract: Vernal keratoconjunctivitis (VKC) is an unusually severe allergic eye disease, occurring mainly in children. Giant papillae are frequently observed on the upper tarsal conjunctiva and expose, sometimes, to severe corneal complications, involving the visual prognosis. Other allergic states, conjunctival staphylococcus aerus and worm intestinal infection, were frequently associated with VKC. We report the case of a 9-year-old girl followed for five years for severe vernal keratoconjunctivitis resistant to treatment with topical corticosteroid. In front of, the high ASLO title, the isolation of pyogenic streptococcus in the throat swab culture, and the presence of a chronic pansinusitis, the diagnosis of a post-streptococcal syndrome was retained. The patient is treated with protected amoxicillin for 12 days. The evolution was spectacular with almost total disappearance of the giant papillae without any recurrence, after 6 months of follow-up. In our knowledge, this case is the first case of vernal keratoconjunctivitis with giant papillae reported as being a post-streptococcal immune complication. Ophthalmologists, confronted to vernal keratoconjunctivitis, should consider post-streptococcal syndrome as a possible cause. The search for an increasing title of ASLO and streptococcal infectious evidence can establishes the association.

Keywords: Vernal keratoconjunctivitis – tarsal giant papillae- Post-streptococcal syndrome- streptococcus.

INTRODUCTION

Vernal keratoconjunctivitis (VKC) is a bilateral, usually seasonally recurrent, allergic inflammation of the conjunctiva, characterized by limbal gelatinous hypertrophy and/or upper tarsal giant conjunctival papillae [1].

Topical corticosteroids are often used during acute flare-ups in VKC [2]. However it's unsatisfactory for controlling severe cases and avoiding recurrences [3].

The possible role of intestinal parasitic infection [4, 5] and conjunctival staphylococcus aerus [6]11in an occurrence and aggravation of these ocular inflammatory diseases, is counterversed.

We report, the first case of vernal kerato-conjunctivitis with giant papillae reported as being a poststreptoccal immune complication and we discuss in a review of the literature the other possible associations with this pathological inflammatory ocular.

CASE PRESENTATION

9- year- old girl. She has been followed for 5 years by her doctor for a vernal keratoconjunctivitis. She has repeatedly received local and general steroids with artificial tears. It is planned for a topical 0.05% cyclosporine.

The patient presented at the 4th week of treatment with regressive topical dexamethasone associated with several artificial tears and a topical antibiotic. It presents a tearing e with redness of the eye and irritability of the conjunctiva.

According to the parents, the child had a history of recurrent tonsillitis, without notion of an atopic disease. At presentation, the best corrected visual acuity was: 8/10 in the right eye and 9/10 in the left eye .The tarsal conjunctiva is the seat of several giant papillae arranged in paving stones (fig1). At the level of the cornea, there is the presence of dry eye with (breakup-time at 6 seconds) and a minimal superficial punctate keratitis. The rest of the exam is normal.

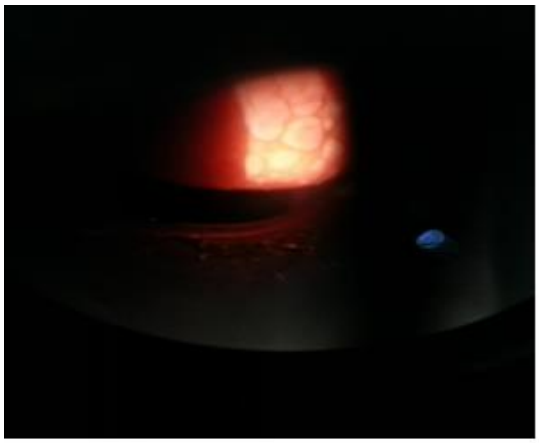

Fig-1: tarsal conjunctivitis with giant papilla in cobblestones

Laboratory tests were applied comprising: complete blood count, sedimentation speed, C-reactive protein, corneo-conjunctival bacteriological sampling, cytobactériology urine, ASLO title with throat swab culture, sputum culture and stool examination. A radiological assessment includes an X-ray of the lungs and a blondoscan

RESULTS

The patient has an inflammatory syndrome with 35mm / 1h and C-reactive protein at 9mg / L. The ASLO title, measured twice at a two-week interval, was 480 / ml and 600 / ml respectively. Swabbing in the throat resulted in pyogenic streptococci. Post-streptococcal syndrome was retained and oral co-amoxiclav treatment was initiated with systemic corticosteroids for 6 days.

The evolution was spectacular with almost total regression of the giant papillae in the tarsal conjunctiva of both eyes (fig. 2).

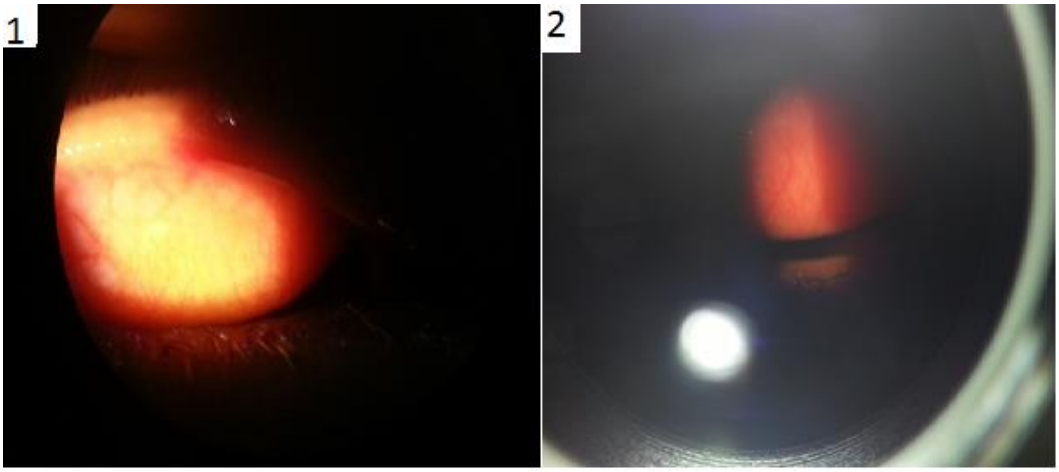

Fig-2: after 10 days of treatment with oral co-amoxiclav.2: evolution after 21 days: showing the disappearance of the giant papillae

Meanwhile the patient was referred to an otolaryngologist who realized a *Scan* of the *sinuses* revealing a polyploidy thickening of the sinus cavities with a thickened part in the mucosal right maxillary sinus, an almost complete filling of the left maxillary and ethmoid sinus some cells, with a lower recess the thickening of the frontal sinuses and sphenoid sinuses. So the patient was retreated by oral co-amoxiclav and steroids (fig.3).

A month later, the giant papillae and sinusitis are despaired. No recurrence was detected after 6 months of follow-up.

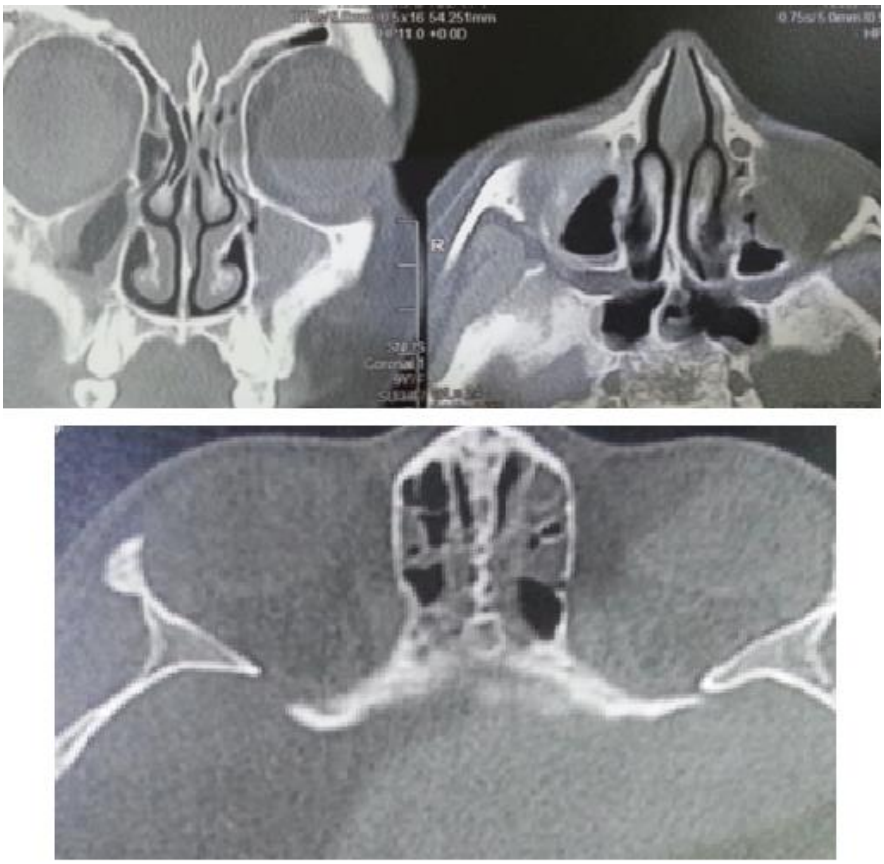

Fig-3: Scan of the sinuses showing a chronic pansinusitis

On the other hand, the control ASLO title remains high at 430 ui / l and the girl is proposed for a tonsillectomy.

DISCUSSION

Vernal keratoconjunctivitis (VKC) is a relatively rare, chronic form of ocular allergy that can potentially cause severe visual complications. Clinically characteristic findings are tarsal giant conjunctival papillae (> 1 mm) and/or limbal gelatinous changes (Trantas dots) [1, 2].

VKC is a disease showing great racial and geographical variation. It is most common and most severe in hot, arid environments such as the Mediterranean basin, West Africa, and the Indian subcontinent [7]. In these areas, up to 3% of eye clinic patients present with VKC and 10% of outpatient appointments are made for signs and symptoms related to VKC [8].

In the conjuctival form- Usually upper tarsal conjunctiva of both the eyes is involved. Typical lesion is characterized by the presence of hard, flat-topped papillae arranged in cobblestone or pavement stone fashion. In severe cases papillae undergo hypertrophy to produce cauliflower-like excrescences of giant papillae [9].

As is the case with our patient, children with VKC present with severe ocular symptoms, that is, severe eye itching and irritation, constant tearing, red eye, eye discharge, and photophobia [1-3].

Topical corticosteroids are often required for the treatment of flare ups [2, 3]. However this management is unsatisfactory in controlling severe cases and avoiding recurrences [2]. Some VKC patients will face sight-threatening complications which are mainly due to corneal involvement and iatrogenic damage caused by prolonged corticosteroid treatment [1, 2].

A family history of allergic diseases occurs in 35.5% of VKC cases, with positivity for other associated allergic states such as asthma, eczema, and/or rhinitis found in 37.1%. A positive skin prick test has been identified in 51.4% of patients [10].

Worm infection is particularly roundworms more is strongly associated with VKC [4] in two series of cases, [11, 12] an anti-parasitic treatment has been reported to improve the evolution of KCV. However, Abhisek Mondal *et al*. [13] rapport that Intestinal parasitic infestation did not play a role in VKC, and treating intestinal parasites will probably not help in the better prognosis of VKC.

Ahmed M. Al – Hakami [14], in their study of 18 VKC patients and 22 healthy controls in order to study the association between the presence of bacteria and the occurrence of VKC, found that Staphylococcus aureus was detected more frequently in VKC (27.78% vs. 4.55% in control, p = 0.041) and Staphylococcus epidermidis were more common in control of the eyes (45.45% in control vs. 5.56% in VKC, p = 0.005) and concluded that S. aureus colonization had an aggravating effect role, in the occurrence of VKC, by contrast. Epidermidis have a possible role of against the occurrence of VKC.

However, Kerr N. and al. [5] reports that bacterial keratitis, in particular staphylococcus aureus, is due to abnormalities of ocular immune mechanisms found in patients with vernal keratoconjunctivitis. Our patient has no notion of allergic disease. Parasitological examination of the stool did not reveal any parasite and the corneo-conjunctival sample was sterile.

In this case, the repeated and unsuccessful treatment by local steroids and eye drops, the inflammatory syndrome associated with the high ASLO title, and the isolation of pyogenic streptococci in the throat swab culture, allows us to retain the diagnosis of a streptococcal infection disease. Resolution of clinical signs confirms previous suspicions about the etiopathogenic role of streptococcus in this conjunctival lesion.

Isolated conjunctival involvement in the form of vernal kerato-conjunctivitis with large papillae in pavement, to our knowledge is never reported in the literature. Only an exceptional case of conjunctival nodule simulating lymphoma [16] is reported until now. Our case is, therefore, the first case of vernal kerato-conjunctivitis with giant papillae reported as being a poststreptoccal immune complication.

Repeated SGA infection may be responsible for several immune-related complications of cardiac, articular, dermatological, renal, cerebral and ocular localization and pediatric autoimmune neuropsychiatric [17].

At the eye level, although bilateral anterior non-granulomatous anterior uveitis is often encountered [18, 19], uveitis post streptococcal syndrome is characterized by a clinical polymorphism. Posterior involvement is reported between 37 and 50% of cases [18, 19] is characterized by a wide variety of non-specific lesions. Other aspects associated with anterior uveitis have been reported but are very rare, such as glaucoma, scleritis [20], and kerateuveitis [21].

Although the exact pathophysiological mechanism of PSS is still unknown, most authors propose a molecular mimicry hypothesis between bacterial antigens and autoantigens leading to autoimmunity [22].

The peptidoglycan-polysaccharide complexes in the wall of streptococcus bacteria have many properties that can activate phagocytic cells and involve the complement cascade to cause tissue injury [23]. The low biodegradability of PG-PS [24] helps to explain how this bacterial debris persists in tissues and act as a chronic stimulus. PG-PS also has an antigenic role, and therefore its chronic presence in the tissue could lead to the production of immune complexes and to an immunopathology [23].

Streptococcal infection has also ability to penetrate epithelium surface causing invasive infection. This invasive form can be severe, or at times moderate and unnoticed. Dendritic cells, macrophages, and T cells that reside in subepithelial tissues recognize bacterial antigens, resulting in the production of Th1-polarized proinflammatory cytokines, including interferon (IFN) -γ and tumor necrosis factor (TNF) -α [25,26]. These cytokines cause an inflamed phenotype of epithelial cells, resulting in the production of host defense molecules, including chemokines [27].

These proinlammatory cyokines derived from th1 and / or th2 lymphocytes resulting from streptococcal bacterial adhesion, are also implicated in the pathogenesis of VKC. In fact, The clear abundance of Th2 cytokines, upregulated expression of their receptors and conspicuous paucity of T helper cell type 1 (Th1) cytokines in tear and serum of VKC patients confirm the crucial role played by these factors in the onset and perpetuation of the chronic allergic inflammation observed in VKC [1].

CONCLUSION

Ophthalmologists, confronted to vernal keratoconjunctivitis, should consider post-streptococcal syndrome as a possible cause. The search for an increasing title of ASLO and streptococcal infectious evidence can establishes the association

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