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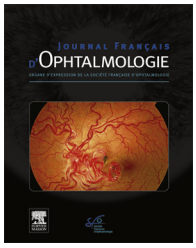

LETTER TO THE EDITOR

Occurence of bilateral keratoconus and basal laminar drusen: A chance association or a true relationship?

Manifestation d'un keratocone bilateral et d'un basal laminar drusen : association hasardeuse ou veritable relation ?

Keratoconus and cuticular drusen (CD) are two uncommon diseases, possibly due to oxidative stress and dysfunction of extracellular matrix [1,2]. We report the first known case of coexistence of bilateral keratoconus and CD and discuss the possible links between the two conditions.

A 21-year-old male patient presented a monocular diplopia of his left eye. His best-corrected visual acuities (BCVA) were 0.1 logMAR (− 0.75 spherical (SPH), − 1.5 cylindrical (CYL) × 155) in his right eye and 0.4 logMAR (− 2 (SPH), − 6 (CYL) × 160) in his left eye.

Slit lamp examination was normal in the right eye and revealed stromal thinning, Vogt's striaes, and corneal nerve hypertrophy in the left eye. The fundus showed the existence of numerous bilateral small hard drusen. Spectral-domain optical coherence tomography (SD-OCT) confirmed the presence of bilateral small hard drusen (Fig. 1 E and F). Fluorescein angiography showed the characteristic ''stars-in-the-sky'' appearance in both eyes (Fig. 1 G and H), suggesting the diagnosis of CD. Ocular response analyzer (ORA) disclosed a bilateral decrease of corneal hysteresis predominantly in the left eye (Fig. 1 A and B), and corneal topography confirmed the bilateral keratoconus (Fig. 1 C and D) with 7.5 dioptres of irregular astigmatism in the central 3 mm in the left eye.

The family history found the notion of bilateral keratoconus with keratoplasty for his father. There was no consanguinity. The father's fundus examination, OCT and the fluorescein angiography were normal.

CD, also termed as ''basal laminar drusen'', is characterized by the fundoscopic findings of innumerable, uniformly sized, small (25 μm to 75 μm) and round drusen. In early phases of fluorescein angiography, these drusen produce a typical ''stars-in-the-sky'' or ''milky way'' pattern. Its pathogenesis involves local inflammation and extracellular deposits [3,4].

Keratoconus is another degenerating disease characterized by central thinning, increased curvature and scarring of the central region of the cornea. The pathogenesis of keratoconus is still unclear, but it has been hypothesized that the tissue degradation is linked with oxidative stress and local inflammation [1,5].

As previously reported [3,6—8], analyses of CD and keratoconus corneas composition have identified many common components. Some biochemical abnormalities identified in keratoconus corneas also exist in CD environment. In fact, some authors reported altered lectin-binding sites in keratoconus corneas, such as Ricinus communis agglutinin I (RCA-I), which is also a constituent of CD [3,6,8]. Moreover, amyloid beta, a major component of CD have previously been detected in some cases of keratoconus [3,7]. We hypothesize that a metabolic alteration of one or more of these common components may induce the co-occurrence of these two conditions.

Many papers supported that local inflammation and oxidative stress are implicated in both CD and keratoconus pathogenesis. Corneal thinning in keratoconus as well as the basal laminar deposits involves the expression of certain inflammatory mediators [1,2]. We hypothesise that a common alteration of a factor related to oxidative stress may explain this association. In fact, it have been reported that interleukin 6 (IL-6), a key factor in the modulation of immune and inflammatory responses, is overexpressed in the tears of keratoconus eyes as well as in aqueous humour of eyes with drusen-related diseases [1,2,5,9]. HLA DR antigens (major histocompatibility complex class II) are another frequent common compound found in both CD and keratoconus [3,10].

Finally, some cases of central serous chorioretinopathy in keratoconus eyes have already been reported [11]. Central serous chorioretinopathy is a retinal condition that, like the basal laminar drusen, is associated to retinal pigment epithelium dysfunction [3,4]. The hereditary pattern for keratoconus occurring in this case may suggest the role of genetic factors, but the father did not have retinal manifestations. To our knowledge, this is the first clinical report of association of bilateral CD and keratoconus. The relationship between these conditions is hypothetical and is still difficult to explain. Further investigations including genetic and biomarkers analysis will be necessary to assess if there

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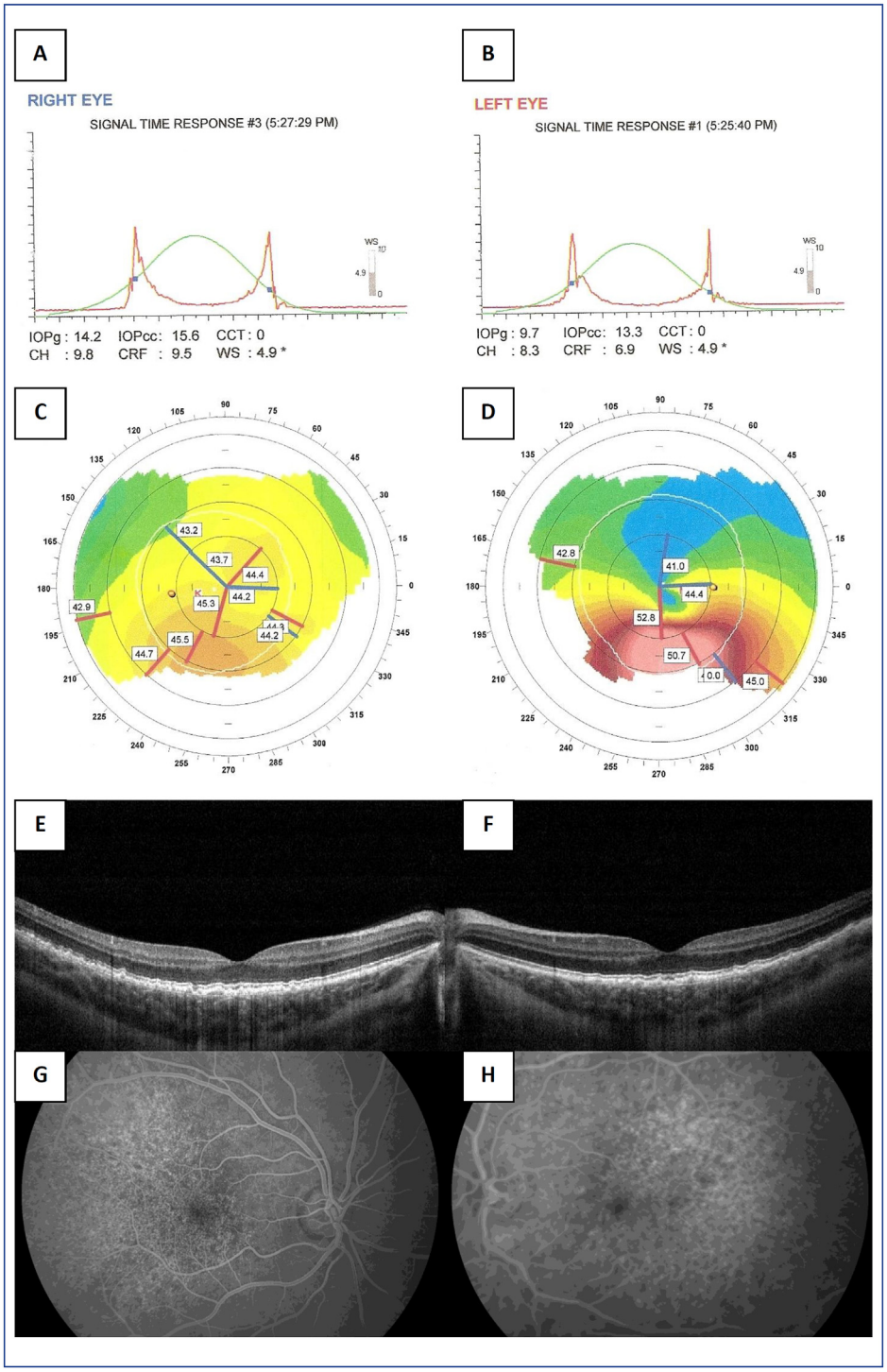

Figure 1. A. Ocular Response Analyzer of the right eye showed a decrease of corneal hysteresis. B. Ocular Response Analyzer of the left eye showed a more important decrease of corneal hysteresis. C and D. Corneal topography showed bilateral keratoconus pattern, which was more important in the left eye. E and F. Presence of bilateral small hard drusen on OCT scans. G and H. Presence of a typical ''stars-in-the-sky'' appearance at the intermediate phase of fluorescein angiography. Note that the image is blurred in the left eye because of keratoconus.

is a possible pathogenic correlation between these two entities or if this is just a chance coexistence of two separate pathologies.

Disclosure of interest

The authors declare that they have no competing interest.

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R. Tahiri Joutei Hassani[a,*,b], G. Querques[c], H. Liang[d,e], E. Souied[c], C. Baudouin[d,e,f,g]

[a] *Department of Ambulatory Surgery, Granville Hospital, Granville, 50400, France*
[b] *Ophthalmology Practice ''Le Visiologiste'', Granville, 50400, France*
[c] *Department of Ophthalmology, Creteil Eye Clinic, University Paris-Est-Creteil, Creteil, 94000, France*
[d] *Department of Ophthalmology III, Quinze-Vingts National Ophthalmology Hospital, Paris, 75012, France*
[e] *Center of Clinical Investigations 503, Quinze-Vingts National Ophthalmology Hospital, Paris, 75012, France*
[f] *Ambroise-Paré Hospital, Versailles St Quentin en Yvelines University, AP—HP, Boulogne-Billancourt, 92100, France*
[g] *Inserm UMRS 968, Vision Institute, University Pierre-et-Marie-Curie, Paris, 75012, France*

* Corresponding author: Department of Ambulatory Surgery, Granville Hospital, 849, rue des Menneries, 50400 Granville, France.
E-mail address: tjhr78@hotmail.com (R. Tahiri Joutei Hassani)

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