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Bilateral conjunctival Splendore-Hoeppli phenomenon associated with juvenile xanthogranuloma

The Splendore-Hoeppli phenomenon is considered a rare and distinct local mucocutaneous inflammatory reaction to antigens of a vast variety of infectious agents including fungi, bacteria, and parasites, and to some foreign material such as silk sutures.[1] Hypereosinophilic syndrome and allergic conjunctival granulomas are noninfectious disorders that may also be associated with Splendore-Hoeppli phenomenon.[1,2] In most conjunctival lesions, however, the definitive causative agent cannot be identified.[2] We describe a case of spontaneously regressed multiple bilateral conjunctival Splendore-Hoeppli lesions in association with juvenile xanthogranuloma of the scalp.

A 6-year-old male was brought by his parents for itchy eyes and small opaque nodules over both eyes that developed within 10 days. Six months previously, a red papular lesion had been removed from his scalp, which proved to be juvenile xanthogranuloma. On our examination, his visual acuity was 20/20 OU. There were bilateral, multiple, small, pale yellow, and irregular conjunctival nodules (Fig. 1). All were located in the superior half of the conjunctiva. The lesions were painless, soft, and mobile on palpation. Dilated episcleral vessels were also observed. The rest of the ocular examination was noncontributory. A full blood count and blood biochemistry tests were normal; no eosinophilia was detected. A week later, an excisional biopsy of 1 of these lesions was performed. However, at the time of the operation most of the lesions had already disappeared bilaterally. No treatment was offered and there has been no recurrence during follow-up.

Histopathologic examination of the specimen showed granulomas beneath the conjunctival epithelium. The granulomas were composed of multinucleated giant cells, lymphocytes, and epithelioid histiocytes surrounding bright eosinophilic amorphous material and cellular debris (Fig. 2A). Splendore-Hoeppli materials were observed inside the giant cells (Fig. 2B). No specific microorganism was identified. A systemic work-up in search of an infectious disease was noncontributory, and the stool tested negative for parasites.

The Splendore-Hoeppli phenomenon is characterized by an amorphous, eosinophilic substance surrounded by multinucleated giant cells, lymphocytes, epithelioid histiocytes, and eosinophils.[1,2] This material is usually composed of either eosinophilic major basic protein or antigen–antibody complexes, possibly deposited against an infectious agent.[1,2] Splendore-Hoeppli phenomenon in a patient with juvenile xanthogranuloma has not been previously reported.

Juvenile xanthogranuloma is a self-limited benign disorder of early childhood characterized by a solitary, red or white cutaneous papule or nodule, and it belongs to non-Langerhans cell histiocytoses.[3] Histopathologically, proliferated histiocytic cells of $CD14^+$, $CD68^+$, $fascin^+$, and factor $XIIIa^+$ dendrocyte phenotype form the lesions.[4] The characteristic lipid-laden Touton giant cells have a macrophage morphology.[4] Juvenile xanthogranuloma has been observed in association with neurofibromatosis types 1 and 2 and juvenile chronic myelomonocytic leukemia, suggesting Ras pathway deregulation as a common pathogenetic basis.[4] Although juvenile xanthogranuloma and Splendore-Hoeppli phenomenon have distinctly separate histopathologic features, further observations are necessary

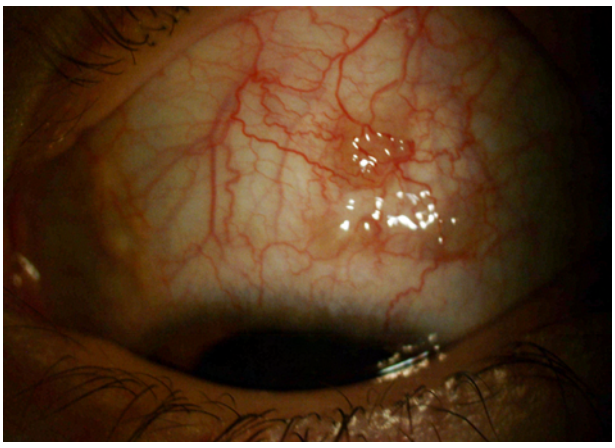

Fig. 1—Biomicroscopic view of the superior conjunctiva of the left eye showing irregular, yellow-white tumefactions.

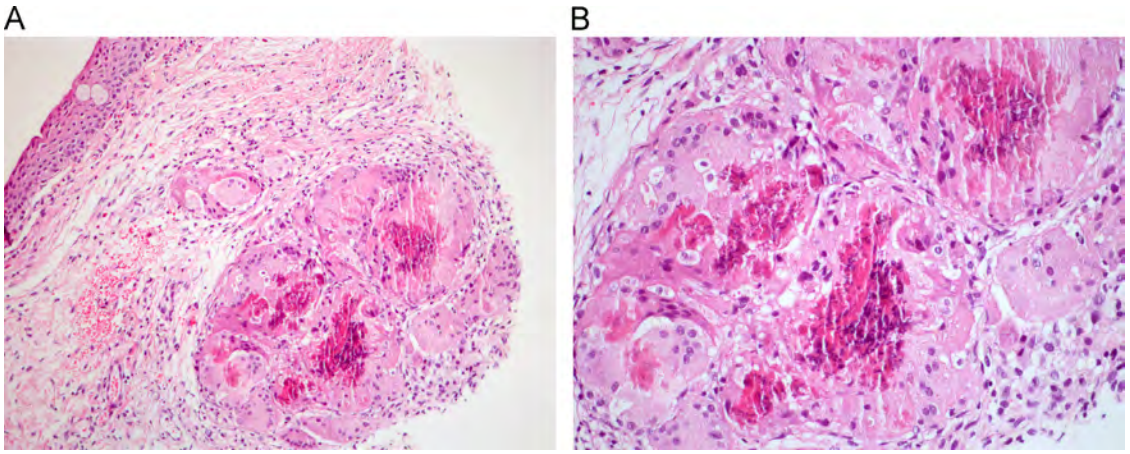

Fig. 2—Histopathologic view of the specimen. A, Strongly eosinophilic amorphous material surrounded with epithelioid cells and multinucleated giant cells (hematoxylin and eosin [HE] staining, original magnification ×100). B, Splendore-Hoeppli material seen inside the giant cells (HE staining, original magnification ×200).

to determine whether these conditions may share common immunopathogenic dysfunction at some point during their evolution. The coexistence of these disorders in our patient may also be merely coincidental.

There is no established treatment for the Splendore-Hoeppli phenomenon. The use of topical or systemic corticosteroids is controversial because this was found to be of no benefit in many patients, as demonstrated in a patient in whom the Splendore-Hoeppli phenomenon developed under high-dose oral steroids and disappeared during withdrawal of the drug.[5] Regression either spontaneously or disappearance of other lesions after surgical excision of 1 lesion was also occasionally observed.[2,6] Recently successful outcome was reported after use of topical cyclosporine A for 18 months.[7] Our experience with the patient with bilateral involvement supports the observations that the Splendore-Hoeppli phenomenon is a self-limited disorder that may completely resolve spontaneously.

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Spontaneous resolution of optic disc pit maculopathy after posterior vitreous detachment

We report the case of a 57-year-old male with optic pit maculopathy that spontaneously resolved after posterior vitreous detachment (PVD). Progressive improvements in visual acuity, clinical examination, and spectral-domain optical coherence tomography (SD-OCT) occurred over 3 months. The pathophysiology of optic pit maculopathy has yet to be completely elucidated. This case report lends additional support to the theory that the vitreous and its attachments at the optic nerve head play a critical role in the development of optic disc pit (OPD) maculopathy (ODP-M), and that the PVD may play a critical role in its treatment.

The prevalence of ODPs is approximately 1 in 11000 persons. Approximately 25% to 75% of eyes will develop a secondary maculopathy, most commonly in early