

Uveitis associated with immunoglobulin E-mediated allergic diseases

ABSTRACT

Atopic dermatitis and food allergy are typically associated with elevated IgE levels. Ocular involvement in atopic dermatitis is usually limited to the ocular surface and ocular adnexae. We herein report on the unusual association of these two conditions with uveitis in two patients and describe the ocular and systemic outcome following the institution of systemic immunosuppression.

Key words: atopic dermatitis, atopy, food allergy, uveitis.

INTRODUCTION

Atopic dermatitis (AD) is a chronic inflammatory skin disease commonly associated with respiratory allergies such as rhinitis and asthma, and is classically associated with a high serum level of IgE.

Food allergy, defined as an adverse immune response to food proteins, is most common in young children, especially among those with AD. Food-induced allergic reactions might be caused by IgE-mediated and non-IgE-mediated mechanisms.

We herein report on the association of these two IgE-mediated conditions with uveitis in two patients and describe the outcome of both the ocular and systemic conditions in response to systemic immunosuppression.

CASE REPORTS

Case 1

A 41-year-old man was referred to the uveitis service at Aberdeen Royal Infirmary hospital because of bilateral recurrent anterior uveitis. Past medical history revealed that he suffered from childhood asthma and mild eczema with no ocular involvement. His allergic manifestations subsided as a teenager albeit relapsing at 32 years of age. At that time he had widespread eczema over his trunk and face with mild conjunctivitis. Serum IgE level was highly elevated at 988 IU/L (range 2–120 IU/L) and he had a strongly positive skin patch test to house dust, dog dandy and moulds. He was initially treated with oral antihistamines and steroid ointments with partial improvement. Within a few months he developed atopic keratoconjunctivitis (AKC) with bilateral corneal infiltrates, corneal vascularization and progressive corneal scarring. Topical therapy with mast cell stabilizers, antihistamines and steroids was of no benefit. Surgical interventions were undertaken for the correction of lower lid cicatricial ectropion, lacrimal duct stenosis and ptosis. At 40 years of age, he suffered from right anterior non-granulomatous uveitis which resolved with topical steroids but which recurred bilaterally 1 year later. On examination visual acuity (VA) was 6/60 right eye and 6/18 left eye. He had severe eczema periorbitally with marked blepharitis and papillary conjunctivitis. He had bilateral corneal pannus with scarring in the inferior parts of the corneas. Bilateral anterior chamber cells (+1) were seen with posterior synechiae. Systemic work-up was unremarkable. Treatment was started with low-dose prednisolone (20 mg/day) and

cyclosporin (5 mg/kg/day) along with topical steroids. He immediately recovered from the uveitis and within few months the AKC and skin disease became completely inactive. His vision significantly improved to 6/9 in each eye following regression of corneal infiltration and vascularization. Since the institution of immunosuppressive therapy, he has had no relapses of AKC, uveitis or eczema during a follow-up period of 11 years. He remains well controlled on low-dose prednisolone and mycophenolate mofetil which was instituted in the last 2 years of his follow up as cyclosporin was discontinued because of mild renal impairment.

Case 2

A 4-year-old boy presented to the uveitis service at Aberdeen Royal Infirmary hospital with bilateral panuveitis. He had been suffering from mild eczema and food allergy since 8 weeks of age. He was well-controlled with oral antihistamines instituted at 2 years of age.

On examination VA was 6/18 right eye and 6/9 left eye. He had small keratic precipitates, moderate anterior chamber cells and flare and posterior synechiae. Posterior segment examination disclosed bilateral inferior snowbanking with peripheral retinal vasculitis. There was bilateral cystoid macular oedema and optic disk swelling, worse in the right eye. Extensive investigations proved negative apart from raised titres of serum IgE and the presence of highly reactive specific IgE to nuts, milk and various other food related allergens. He was put on a strict nut and milk-free diet accordingly with improvement of his general health.

Because of his bilateral active ocular disease, oral cyclosporin (5 mg/kg/day) and prednisolone (1 mg/kg/day) were introduced with good control of the uveitis. Resolution of the bilateral macular, optic disc oedema and vitritis was achieved. In addition, there was restoration of happy temperament. Medications were eventually stopped after 6 years with no more relapses of uveitis for more than 7 years.

Milk was gradually reintroduced in the patient's diet at age of 7 years while a peanut challenge test performed at age of 14 years still elicited an immediate reaction with urticarial rash after antigen application over his lips.

Unfortunately amblyopia developed in his right eye (VA 6/36) because of anisometropic astigmatic myopia ($-3.5 + 1.50 \times 35^\circ$). Left VA was 6/6.

DISCUSSION

Atopic dermatitis is a chronic, highly pruritic, inflammatory skin disease that frequently precedes the development of asthma or allergic rhinitis. It is the most common chronic skin disease of young children, but can affect patients of any age.

The eyelids commonly are involved, and the patients may lose lashes or eyebrows owing to the scratching and rubbing. Conjunctivitis, punctate keratopathy and in advanced cases deep corneal opacities have been described. Herpetic eye infections, keratoconus, cataracts and retinal detachment have also been observed.

Cyclosporine is used successfully in the treatment of severe AD in children and adults, producing a rapid and highly significant improvement in terms of disease activity, pruritis and sleep disturbance.¹

The first patient here was suffering from AD since childhood. From the ocular point of view, he initially suffered of its cicatricial complications on the ocular adnexae and ocular surface. His first

presentation with uveitis was at 40 years of age. Introduction of cyclosporin in addition to prednisolone resulted in resolution of his systemic and ocular manifestations with marked regression and improvement of the ocular surface as well as the uveitis.

The only reported case in the literature on the association of uveitis with AD was in a 15-year-old boy who had been suffering from pruritis since the age of 6 months, and whose presentation was with generalized erythroderma. He had bilateral cataracts, pigmented keratic precipitates and Vossius ring in one eye indicating iridocyclitis. There was no mention of serum IgE level.²

Food allergy is most common in young children. The adverse reaction typically occurs within minutes to a few hours after ingestion and generally involves the dermatologic, gastrointestinal and respiratory systems. Once the diagnosis is established, the only proven therapy is the strict elimination of the food from the patient's diet.

The second patient here suffered from pars planitis and peripheral retinal vasculitis. Remission followed the introduction of cyclosporin. Proper management of his food allergy could have contributed to his favourable outcome. It is not clear to us whether this is a coincidental occurrence or pathogenetic association. One previous report described common foods to be the cause of uveitis in two patients. They obtained marked relief following elimination of these foods from the diet.³

Atopy is regarded as a hyperreactive state of the immune system, with a shift in the T-cell response towards a Th2 dominated activity. Judging from the cross-regulatory properties of Th1 and Th2 cells, one would hypothesize that the prevalence of atopic diseases would be low in Th1 autoimmune diseases. Several reports are showing coexistence of Th2 diseases such as asthma with Th1 conditions such as coeliac disease, insulin-dependent diabetes mellitus and rheumatoid arthritis.⁴ Bilateral optic neuritis was recently described in a patient with strong atopic diathesis and markedly elevated serum IgE level.⁵

Our report here presents an uncommon association between two Th2 diseases namely AD and food allergy that are IgE-mediated and uveitis which has been shown to be predominantly Th1 mediated. Remission was achieved with the use of prednisolone and cyclosporin which is also known to have favourable outcome in treating patients with atopic diathesis.

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Received 15 January 2007; accepted 5 April 2007.

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A case of Oguchi's disease with diabetic retinopathy

ABSTRACT

A 50-year-old Iranian diabetic woman with night blindness was referred to our clinic. Complete ophthalmic examination including ophthalmoscopy after dark adaptation and paraclinic evaluations such as fluorescein angiography and electroretinography were performed. Retinal neovascularization and preretinal haemorrhages compatible with high-risk characteristic proliferative diabetic retinopathy were observed bilaterally. In addition, a golden-yellowish discoloration of posterior pole was noted in her both eyes. The diagnosis of Oguchi's disease was made when this discoloration disappeared after dark adaptation for 3 h. Electroretinograms also confirmed the diagnosis by showing a slow negative wave followed by a slow positive wave in the photopic condition and absent a- and b-waves in the scotopic state. We concluded that proliferative diabetic retinopathy might occur in a patient with Oguchi's disease. This report represents this association in an Iranian patient for the first time.

Key words: diabetic retinopathy, Mizuo–Nakamura phenomenon, Oguchi's disease.

CASE REPORT

A 50-year-old woman with a 15-year history of insulin-dependent diabetes mellitus was referred by her endocrinologist for ophthalmic examination. She had a history of being mildly night-blind all her life and complained of recent blurred vision. Complete ophthalmologic examination including funduscopy after dark adap-

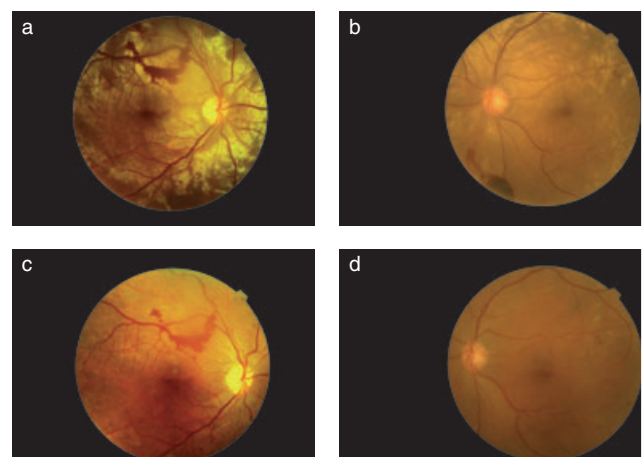


Figure 1. (a), (b) Fundus photographs in normal illumination; (c), (d) after 3 h eye patching.