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Sir,

Corneal Keloid: Aetiology and Management in Lowe's Syndrome

Keloids are hypertrophic scars which result from excessive collagen deposition and most commonly occur in the skin following trauma.¹ Keloid formation in the cornea is rare and tends to occur following ocular injury or perforation,²⁻⁵ but has also been reported in association with rubeola⁶ and Lowe's syndrome.⁷ Congenital corneal keloid has also been described.^{2,5,8,9}

Case Report

A 13-year-old boy with Lowe's syndrome developed bilateral corneal keloids. Lowe's syndrome (oculocerebrorenal syndrome)¹⁰ is an X-linked recessive disorder consisting of renal abnormalities with aminoaciduria, proteinuria and renal tubular acidosis in association with mental and psychomotor retardation, and a characteristic facial appearance. It is associated with cataract formation and there is a high incidence of congenital glaucoma.

The patient underwent bilateral lensectomy for congenital cataracts within the first 3 months of life and was successfully fitted with soft contact lenses. Further surgery to enlarge the pupillary axis of the left eye was undertaken at the age of 6 years, and visual acuities stabilised at 6/60 with soft contact lenses. By the age of 11 years, peripheral corneal opacity had developed in the left eye, and progressively enlarged over a 2 year period to form an elevated, glistening, white mass on the inferior cornea (Fig. 1). Similar changes occurred to a lesser extent in the right eye. The appearances were consistent with keloid formation of the cornea.

Discussion

Corneal keloids tend to present with diffuse involvement of the corneal stroma, or as localised nodules which slowly enlarge, and mainly occur following trauma.²⁻⁷ The nature of corneal keloid formation remains obscure. It may originate in the corneal stroma,^{2,3,5} or could be associated with incarcerated iris.⁴

A continuity between blood vessels arising from a corneal keloid and vessels in the iris has been reported in association with iris pigment granules in the corneal scar, suggesting that the keloid may have originated from iris tissue.⁴ Excessive proliferation of fibrovascular connective tissue during the healing phase of an inflammatory reaction or perforating injury of the eye may result in keloid formation, and iris incarceration in a wound may be a stimulatory factor.³

However, in a report of 4 cases of corneal keloid, it was shown that the principal blood supply to the keloid was from relatively normal peripheral cornea and not iris, with iris pigment present only in 1 traumatic case, suggesting that corneal keloid formation resulted from the process of corneal healing.⁵ Similarly, an ultrastructural study of a corneal keloid resulting from a non-perforating injury suggested that keloid formation was a result

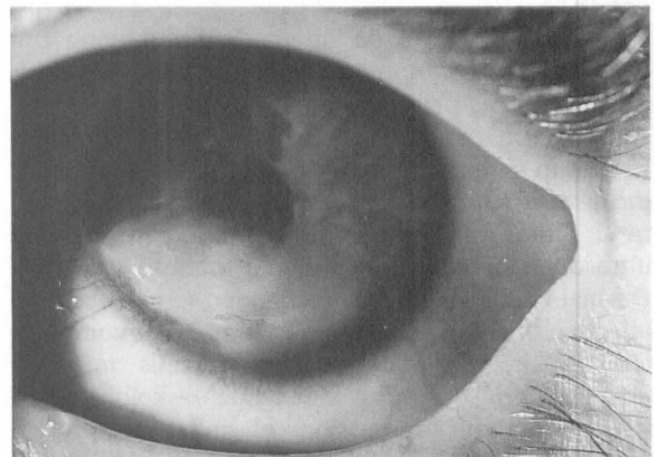


Fig. 1. Keloid affecting the inferior cornea of the left eye.

of the corneal reparative process, without direct involvement of iris tissue.¹¹

In a recent report of 2 cases of bilateral keloid-like myofibroblastic proliferation of the cornea, without a history of trauma or chronic inflammation, it was proposed that previously unrecognised corneal infection could introduce microbial antigens, to invoke an immune-mediated reaction resulting in a hypertrophic scar.⁹

The common occurrence of bilateral corneal keloids in Lowe's syndrome⁷ suggests that in these patients keloid formation may be related to the underlying systemic abnormality. Although cataract and glaucoma surgery is commonly undertaken in such children, routine surgical procedures have not been implicated in keloid formation.

It has been suggested that amino acids may leak into the cornea from abnormal new blood vessels, thus stimulating keloid formation. Substances from the anterior chamber may also leak through defective endothelium. Tripathi and co-authors¹² described bilateral corneal keloid in an 11-year-old boy with Lowe's syndrome who had previously undergone goniotomy to both eyes, and noted that keloid formation had developed in areas not traumatised by the procedure, with no histological evidence of surgical trauma in the area of keloid formation. Similarly, in our case, bilateral corneal keloids developed many years following surgery, with lesions located inferiorly and not related to the site of previous surgery.

The management of corneal keloid poses many problems. Surgical excision of keloids often results in recurrence and enlargement of the lesions.¹ Although essentially benign, keloids may severely affect vision, and large hypertrophic scars can interfere with lid closure.

The deep stromal involvement of the lesions precludes lamellar keratoplasty. Equally, penetrating keratoplasty in these young and mentally retarded individuals may not be considered a viable option.⁸

Treatment should be directed at removing the existing lesion and preventing recurrence by inhibiting fibroblastic proliferation and collagen synthesis. The use of radiotherapy in the management of keloids in general has been reported – in particular, the use of superficial X-rays or strontium-90 beta-rays. Other physical forms of treatment include ultrasound, cryotherapy, pressure therapy and laser. Dermal keloids have also responded to intralesional injection of steroid.¹ Cibis and co-authors⁷ commented on the persistence of mast cells in their ultrastructural analysis of corneal keloid, and suggested that suppression of inflammation by topical steroid or the use of mast cell stabilisers may be useful in the management of this condition.

The management of corneal keloids should therefore be conservative in the first instance, with surgical intervention reserved for those lesions which severely affect vision or impede adequate lid closure.

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Sir,

Localised Giant Papillary Conjunctivitis Secondary to a Dermolipoma

Dermolipomas are solid choristomas formed from displaced embryonic material, destined to become skin, that has been sequestered to the conjunctiva.¹ They typically occur on the superolateral epibulbar surface with a posterior extension into the orbit.² The appearance is of a smooth pinkish-yellow mass in the conjunctiva. Histologically they consist of a keratinised squamous epithelium containing adnexal