

a site for entry of pathogens. The concurrent use of systemic prednisolone may have induced an immunocompromised state, increased her susceptibility to infection, and caused delayed wound healing leading to the late onset of infection.

Although intralesional corticosteroid remains the treatment of choice and is generally safe, clinicians should still be wary of the potential morbidity associated with this procedure.

### Acknowledgements

No financial support was received for this paper.

### References

- 1 Haik B, Karcioğlu Z, Gordon RD, Pechous BP. Capillary hemangioma (infantile periocular hemangioma). *Surv Ophthalmol* 1994; **38**: 399–426.
- 2 Wasserman BN, Medow NB, Homa M, Hoehn ME. Treatment of periocular capillary hemangiomas. *J AAPOS* 2004; **8**(2): 175–181.
- 3 Sutula FC, Glover AT. Eyelid necrosis following intralesional corticosteroid injection for capillary haemangioma. *Ophthalmic Surg* 1987; **18**(2): 103–105.
- 4 Glatt HJ, Putterman AM, Van Aalst JJ, Levine MR. Adrenal suppression and growth retardation after injection of periocular capillary haemangioma with corticosteroids. *Ophthalmic Surg* 1991; **22**(2): 95–97.
- 5 Brooks SE, Pillai JJ, Bains H. Orbital cellulitis due to occult orbital lymphangioma. *South Med J* 2001; **94**(5): 532–535.

LP Ang<sup>1,2</sup>, M-W Lee<sup>1</sup>, L-L Seah<sup>1</sup>, P Cheong<sup>1</sup> and J Rootman<sup>3</sup>

<sup>1</sup>Singapore National Eye Center, Singapore, Singapore

<sup>2</sup>Department of Ophthalmology, National University of Singapore, Singapore, Singapore

<sup>3</sup>Department of Ophthalmology & Visual Sciences and Pathology, University of British Columbia and the Vancouver General Hospital, Vancouver, British Columbia, Canada  
E-mail: leopk@pacific.net.sg

Proprietary interests: The authors have no proprietary interests in any drugs and equipment described in this paper

*Eye* (2007) **21**, 999–1001; doi:10.1038/sj.eye.6702796; published online 30 March 2007

Sir,  
**Peripheral ulcerative keratitis in pityriasis rubra pilaris**  
Pityriasis rubra pilaris (PRP) is an idiopathic, inflammatory, papulosquamous disease characterised by keratotic follicular papules, reddish-orange scaly erythroderma, and palmoplantar keratoderma. The skin, nails, mucous membranes, and eyes can be affected.<sup>1</sup> Histopathology shows follicular plugging, acanthosis, hyperkeratosis, and a lymphohistiocytic

dermal infiltrate.<sup>2</sup> Treatment includes emollients, systemic steroids, retinoids, and immunosuppressants.<sup>3</sup>

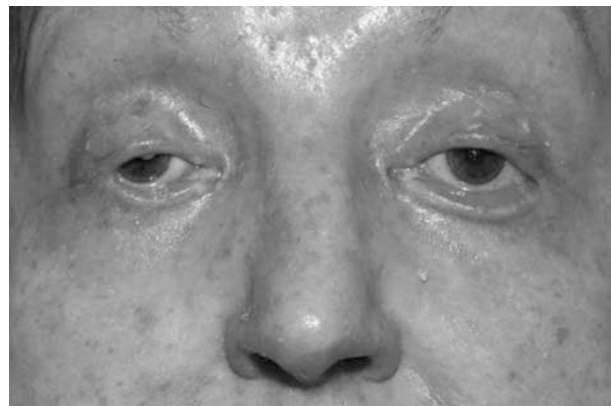
Among the documented ocular signs and symptoms are ectropion and dry eyes (Figure 1). Duke-Elder<sup>4</sup> described changes including connective tissue invasion of Bowman's zone, linear streaks on the cornea, interstitial keratitis, epithelial erosions, and conjunctival keratinisation in PRP. To our knowledge, peripheral ulcerative keratitis (PUK) has not been reported in association with PRP. This report describes a patient with PRP who presented with a possible PUK complicated with a perforation, requiring tectonic penetrating keratoplasty.

### Case report

A 69-year-old female patient presented with a 3-day history of a 'foreign body' sensation in the left eye. This was noticed following minor trauma, when she had hit her left eye with the leg of her spectacles.

On examination, she had bilateral ectropion. Her visual acuities were 4/9 OD and 4/60 OS. In the left eye, there was a corneal perforation measuring 2 × 1 mm at the inferior mid-periphery plugged by iris tissue. There was no epithelial defect or apparent infiltration beyond the margin of the perforation. Siedel test was negative with a quiet, formed anterior chamber and intraocular pressure of 11 mm Hg. This patient was diagnosed with PRP in 2002 and had been on systemic steroids and acitretin for 3 years. She previously had a right tarsorrhaphy and was on regular ocular lubricants.

An urgent tectonic penetrating keratoplasty and lateral tarsorrhaphy were performed. Corneal histopathology including HSV-1 immunocytochemistry revealed no evidence of an infectious aetiology. A remarkable feature was the absence of Bowman's membrane between the site of perforation and the inferior margin of the cornea (Figure 2). There was associated epithelial thickening and basement membrane irregularity. Inflammatory cells were seen scattered in the vicinity of the perforation and elsewhere in the epithelium. These changes appeared to be a pre-existing abnormality, which may have predisposed to corneal perforation following the trivial injury.



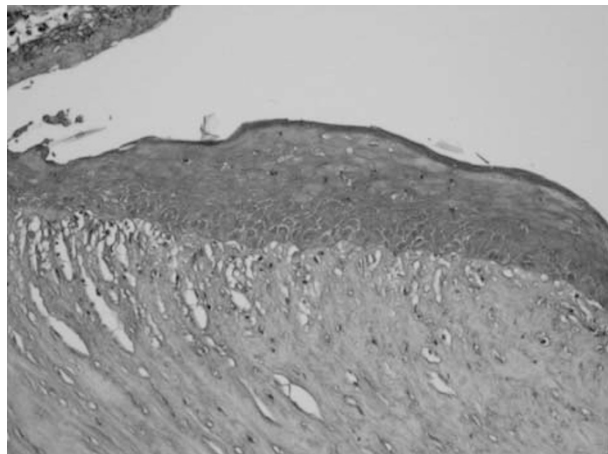
**Figure 1** External picture of patient with PRP, with facial erythroderma and ectropion.

At presentation, her PRP was inactive but flared up a week later. Eight weeks post-operatively, she developed a 90% deep corneal ulcer at the graft–host junction (Figure 3) that remained unchanged in spite of intensive topical lubrication and steroids for 9 days. This ulcer healed rapidly (in 8 days) following the introduction of infliximab, administered by the dermatologists for a concurrent flare-up of PRP.

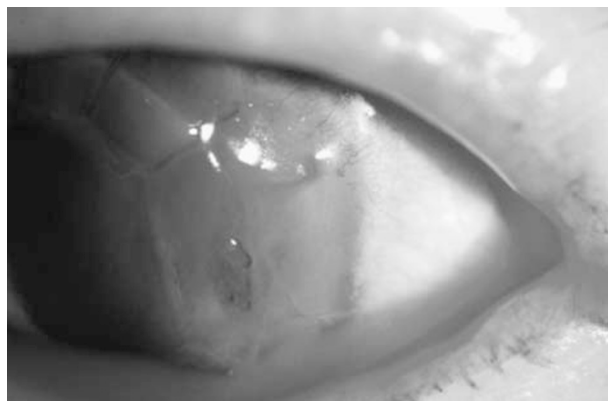
**Comment**

In this patient with PRP, the PUK appeared to be a result of the systemic inflammatory condition, although her ectropion may have contributed to the corneal ulceration. It is unclear whether acitretin, a retinoid used to treat various dermatoses, could be partially responsible.

Similar corneal manifestations in association with inflammatory dermatoses have been described elsewhere in the literature.<sup>5,6</sup> Of particular interest is the absence of Bowman’s membrane on histopathology. There is only one mention of similar findings in the literature. This is in ectodermal dysplasia, a dermatosis with autoimmune aetiology and rare corneal manifestation.



**Figure 2** Corneal specimen after corneal graft in patient with PRP (Masson trichrome, objective magnification ×20) shows thickening of the epithelium and absence of Bowman’s membrane adjacent to the perforation.



**Figure 3** 90% deep corneal ulcer at the graft–host junction.

In this paper, the authors mention PRP as a differential diagnosis.<sup>7</sup>

The corneal ulcer that developed post-operatively improved after the introduction of infliximab, a chimeric monoclonal antibody that neutralises the proinflammatory cytokine, tumour necrosis factor- $\alpha$  (TNF- $\alpha$ ). TNF- $\alpha$  stimulates matrix metalloproteinases that participate in the dissolution of corneal epithelial basement membrane and stroma. Infliximab is used in various inflammatory conditions and has recently been used successfully to treat rheumatoid arthritis-associated PUK resistant to conventional treatment.<sup>8</sup>

In conclusion, we would like to bring to attention the possibility of corneal disease in the form of PUK in PRP.

**Acknowledgements**

We thank Dr Karen Blessing, Western Infirmary, Glasgow, for reviewing the skin biopsy.

**References**

- 1 Allison DS, El-Azhary RA, Calobrisi SD, Dicken CH. Pityriasis rubra pilaris in children. *J Am Acad Dermatol* 2002; **47**(3): 386–389.
- 2 Soeprono FF. Histologic criteria for the diagnosis of pityriasis rubra pilaris. *Am J Dermatopathol* 1986; **8**: 277–283.
- 3 Dicken CH. Treatment of classic pityriasis rubra pilaris. *J Am Acad Dermatol* 1994; **31**(6): 997–999.
- 4 Duke-Elder S. Diseases of the outer eye. In: Duke-Elder S (ed). *System of Ophthalmology*, vol. 8, part 1 C.V. Mosby: Conjunctiva, St Louis, 1965; 560.
- 5 Merchant A, Zhao TZ, Foster CS. Chronic keratoconjunctivitis associated with congenital dyskeratosis and erythrokeratoderma variabilis. Two rare genodermatoses. *Ophthalmology* 1998; **105**(7): 1286–1291.
- 6 Boss JM, Peachey RD, Easty DL, Thomsitt J. Peripheral corneal melting syndrome in association with psoriasis: a report of two cases. *BMJ (Clin Res Ed)* 1981; **282**(6264): 609–610.
- 7 Wilson II FM, Grayson M, Pieroni D. Corneal changes in ectodermal dysplasia. Case report, histopathology, and differential diagnosis. *Am J Ophthalmol* 1973; **75**(1): 17–27.
- 8 Thomas JW, Pflugfelder SC. Therapy of progressive rheumatoid arthritis-associated corneal ulceration with infliximab. *Cornea* 2005; **24**(6): 742–744.

B Paranjothy<sup>1</sup>, M Shunmugam<sup>1</sup>, J MacKenzie<sup>2</sup> and A Azuara-Blanco<sup>1</sup>

<sup>1</sup>Department of Ophthalmology, Grampian University Hospitals NHS Trust, Aberdeen, UK  
<sup>2</sup>Department of Pathology, Grampian University Hospitals NHS Trust, Aberdeen, UK  
 E-mail: manoshun@gmail.com

Meeting presentation: none  
 Financial support: none

*Eye* (2007) **21**, 1001–1002; doi:10.1038/sj.eye.6702797; published online 30 March 2007