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Sir,

In vivo confocal microscopy in congenital rubella keratopathy

Systemic manifestations of congenital rubella syndrome (CRS) include ocular complications (88%), hearing loss (71%), neurologic deficits (75%), and cardiac anomalies (38%).[1] Recognition of ocular manifestations is important in making the right diagnosis in patients with late presentation or without full-blown systemic involvement.[1,2] Cataracts, microphthalmia, glaucoma, and 'salt-and-pepper' retinopathy were the major ocular features, while rubella keratopathy is less common which might be the sequel of either associated glaucoma or direct viral cytopathologic effect.[2] Recent advent of confocal microscopy in delineating corneal pathologies has enchanted *in vivo* analysis of many corneal diseases at cellular level.[3,4]

Case report

A 20-year-old Chinese man without remarkable paediatric history except sensorineural deafness presented to us in July 2002 with bilateral blurring of vision. Best-corrected visual acuities for the right and left eyes were 20/200 and 20/70, respectively. Intraocular pressure (IOP) was 12 mmHg in each eye. Slit-lamp examination revealed diffuse corneal guttata with 'beaten-bronze' appearance. A faint haziness at the stroma was noted but neither Haab's striae nor buphthalmos could be seen. The iris, lens, iridocorneal angles, and corneal sensations were intact. Fundus examination revealed bilateral pigmentary retinopathy and a small juxtafoveal choroidal neovascularization membrane in the left eye (Figure 1a and b). The optic discs and retinal vessels were unremarkable. Small decrement in amplitudes of both scotopic and flicker electroretinogram were noted, suggestive of secondary combined rod and cone involvements. Specular microscopy showed decreased endothelial counts (667 and 712 per square mm in right and left eyes, respectively). Rubella antibody screening by enzyme immunoassay confirmed positive rubella IgG antibody. The loss of hearing, pigmentary retinopathy in this case has already fulfilled World Health Organization recommended case definition for clinically confirmed CRS despite lack of IgM confirmation or other systemic features.[5]

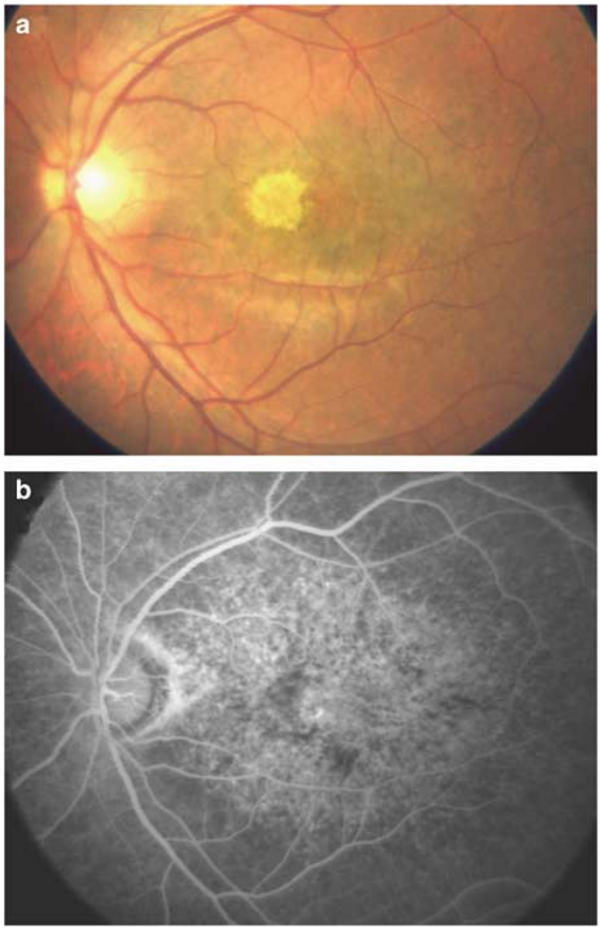

Figure 1 (a) Colour fundus photography of the left eye. It shows a whitish subretinal fibrotic scar at the macula and generalized pigmentary changes at the posterior pole. (b) Fluorescein angiography of the left eye. Hyperfluorescence mottling at the posterior pole is caused by the retinal pigment epithelium atrophy.

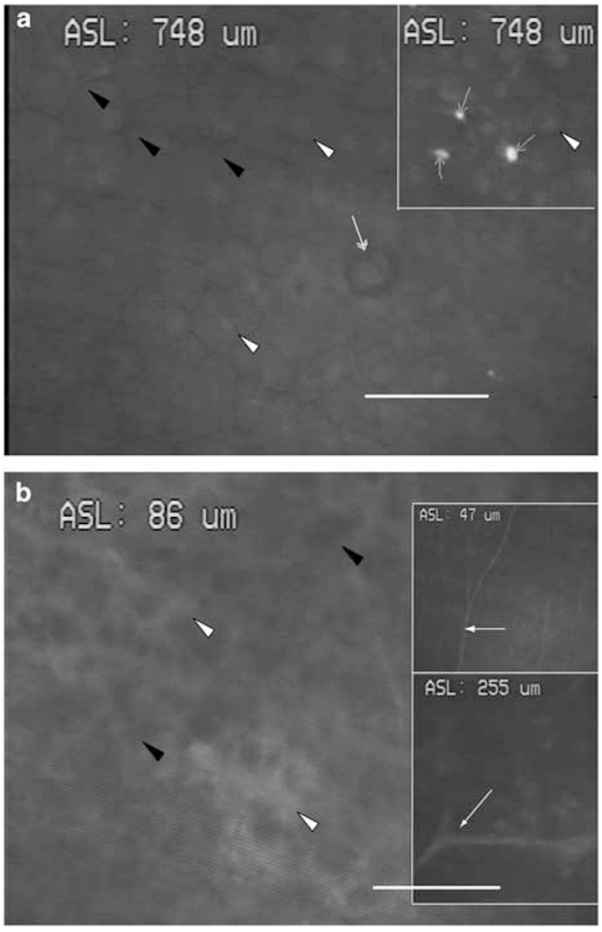

Figure 2 (a) Confocal micrographs of the right corneal endothelium at the depth of 748 μm from the corneal epithelial surface. Large endothelial cells are seen with marked pleomorphism and polymegethism. Most cells lose their characteristics hexagonal cellular border. Nuclei are easily seen (white arrow head). Usual tight appositions of adjacent cellular borders are lost in some areas, leaving a large intercellular space (black arrow head). A 'hollow-out' lesion is found, like an endothelial cell in shrinkage (double head arrow). Insert shows highly reflective, bright lesions in cluster (small white arrow). Inflammatory cell infiltration is not prominent in this micrograph. (Magnification $\times$ 333, untreated photos (except the insertion of illustrating arrows and bars.) White scale bars = 100 μm). (b) Confocal micrographs of the left anterior stroma. Scar tissues were more evident and were shown as dense, well-delineated fibrous tissue in the matrix background (white arrow head). Microcystic spaces (black arrow head) are numerous and prominent. Insets show subepithelial nerve plexus and the deeper stromal nerve fibre trunks, which were comparable to normal cornea (small white arrow). (Magnification $\times$ 333, untreated photos (except the insertion of illustrating arrows and bars.) White scale bars = 100 μm).

Confocal microscopy (ASL1000-ModelOS-1, New Orleans, USA) was performed to evaluate for the cause of corneal oedema in the absence of high IOP. The most striking changes lay in the layer of corneal endothelium (Figure 2a). Significant drop in endothelial cell count were observed accompanied by obvious pleomorphism, polymegethism and increased intercellular gaps. Dark endothelial structures with a central bright reflex 'hollow-out' lesion might signify a cell dying process or a result of direct viral cytopathy. Presence of intracellular microdeposits might further accentuate this supposition as they may represent endothelial viral inclusion bodies. Anterior stromal examination showed microcystic spaces, delineated fibrous tissue, and relatively normal nerve plexus (Figure 2b).

Deluise *et al*[6] have reported two categories of corneal oedema of different aetiologies in congenital rubella syndrome. The one associated with congenital glaucoma tended to be persistent whereas the corneal oedema in the absence of congenital glaucoma was usually focal with spontaneous resolution within 3–6 months.[6] The clinical description of persistent corneal oedema without *sine qua non* of congenital glaucoma was exceedingly unusual throughout English literatures.[6]

Patients with CRS may exhibit progressive disease in which cataract, glaucoma, or keratopathy might manifest them later in life.[7] Confocal microscopic examination is a noninvasive, superbly reproducible instrument to allow real-time *in vivo* assessment of the cornea at cellular level.[3,8–10] The distinctive morphological changes at the endothelium, including cellular irregularities, prominent nuclei, abundant intercellular space, and bright intracellular deposits, advocate the primary disease event residing at endothelium, which can cause subsequent corneal oedema in patients even without IOP elevation.[4,9] These unreported and characteristic *in vivo* corneal morphological changes may fortify our understanding on rubella keratopathy.

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Sir,

Churg–Strauss syndrome in association with proliferative retinopathy

Churg–Strauss syndrome, also known as allergic granulomatosis, was first described by Churg and Strauss in 1951.[1] It is a systemic allergic disease characterised by eosinophil-rich and granulomatous inflammation involving the respiratory tract, and necrotising vasculitis affecting small to medium-sized vessels. The skin, heart, and gastrointestinal tract are occasionally involved, but ocular involvement is unusual. We present the case of a 54-year-old woman who presented with an aggressive unilateral proliferative retinopathy and was subsequently diagnosed as having Churg–Strauss syndrome.

Case report

A 54-year-old female presented to the Ophthalmology department with a 2-week history of hazy vision in her right eye. There was no past ocular history of note. She had a past medical history of hypothyroidism and asthma. Regular medications included thyroxine and salbutamol and becotide inhalers. She smoked 20 cigarettes per day.

On examination visual acuities were 6/24 in the right eye and 6/5 in the left eye. There was a moderate vitreous haemorrhage in the right eye. The left vitreous was clear. There were poor views of the right fundus, but no tears, holes or detachments were seen. The left fundus was healthy. Blood pressure and blood sugar were normal.

The patient was followed up weekly at the retinal out patient clinic. At 3 weeks following her initial attendance the view of the fundus in the right eye had resolved sufficiently to reveal superotemporal new vessels. Fluorescein angiography confirmed the presence of these new vessels (Figure 1) and demonstrated widespread capillary closure (Figure 2).

The patient subsequently underwent right panretinal photocoagulation. Initially, the new vessels appeared to regress. However, 4 months following panretinal photocoagulation she experienced a further vitreous haemorrhage in the right eye. On this occasion the vitreous haemorrhage failed to clear, and therefore 2 months later right vitrectomy combined with endolaser was performed.

The patient was followed up every 2 months in the retinal out patient clinic. At 1 year following her initial presentation, the patient attended clinic complaining of significant systemic symptoms. For the preceding 2 months she had been experiencing dizzy spells and headaches behind her right eye. She complained of lethargy, poor appetite, and had lost a stone in weight. She had noticed progressive numbness in her hands and feet and had developed a nasal discharge. She also

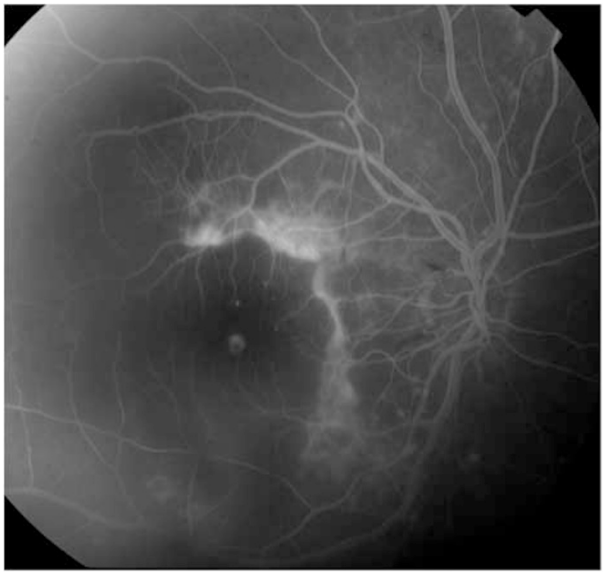

Figure 1 Fluorescein angiography showing superotemporal neovascularisation.