

Anupama Pherwani

Division of Ophthalmology and Visual Sciences,
University of Nottingham, Queens Medical Centre,
Nottingham, UK

Vipul Vakil

PBMA'S H V Desai Hospital, Pune, India

**Habibullah Eatamadi, Ravinder Singh,
Harminder S Dua**

Division of Ophthalmology and Visual Sciences,
University of Nottingham, Queens Medical Centre,
Nottingham, UK

Correspondence to: Professor H S Dua, Division of
Ophthalmology, B Floor, Eye ENT Centre, Queens
Medical Centre, University Hospital, Nottingham. NG7
2UH, UK; harminder.dua@nottingham.ac.uk

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A novel technique to treat traumatic corneal perforation in a case of presumed brittle cornea syndrome

The management of corneal perforation can be difficult. We describe a novel technique to manage corneal perforation in brittle cornea syndrome (BCS).

Case report

A 14-year-old daughter of consanguineous Pakistani parents presented with a history of a bottle cap having struck her left eye. She had a history of multiple trauma to both eyes since childhood.

At presentation, there was limbus-to-limbus corneal perforation in the left eye. The right eye

had a failed corneal graft (for extensive corneal opacities). An examination under general anaesthesia showed a collapsed left eye with rolled-in corneal edges, without any scleral injury. There was partial aniridia, aphakia and prolapsed vitreous, with no obvious retinal detachment. She was also noted to have hypermobility of the small joints of her hands, with bilateral camptodactyly of the fifth fingers and over-riding toes.

During primary repair, the cornea was noted to be soft, with cheese-wiring of 10/0 nylon sutures. At review on post-operative day 1, she had a flat anterior chamber, necessitating a second operation. During this operation, suture track leaks were observed with the use of fluorescein. These were successfully tamponaded with air. To allow sufficient duration of tamponade, a non-expansile (14%) perfluoropropane (C₃F₈) gas exchange was performed after transcorneal three port vitrectomy to gain maximal gas fill. Postoperatively, the cornea was Siedel negative. The patient was kept supine for 10 days, at the end of which the globe remained formed (fig 1).

A month later, the patient received a penetrating corneal injury to the right eye while not using her protective goggles. She underwent primary surgical repair similar to the left eye using C₃F₈ gas. The final visual acuity was hand movement by both eyes.

Discussion

The patient's history suggested an underlying connective tissue disorder affecting the eyes.

BCS is a generalised connective tissue disorder characterised by corneal rupture, after a minor trauma, or spontaneously.¹ Other features include keratoconus or keratoglobus, blue sclera, red hair, hyperelasticity of the skin without excessive fragility, and hypermobility of the joints.² BCS has been reported mainly in Middle Eastern consanguineous families, although no underlying genetic defect has been identified to date.³

A differential diagnosis is Ehlers-Danlos syndrome type VI, which is associated with kyphoscoliosis and thin sclera (with rupture after trivial trauma). It is characterised by the absence or mutation of the procollagen lysyl hydroxylase gene on chromosome 1, causing a deficiency of the enzyme lysyl-hydroxylase.⁴ This leads to a build-up of urinary hydroxylysyl-pyridinoline. In contrast, BCS has normal total urinary pyridinoline ratios. The urinary test results of this patient showed a normal ratio of total lysyl-pyridinoline to total hydroxylysyl-pyridinoline, suggesting a diagnosis of BCS.

Repair of corneal perforations using tissue adhesives and viscoelastic agents,⁵ and onlay epikeratoplasty with a donor corneoscleral button to repair a ruptured keratoglobus,⁶ has been reported previously. Air tamponade is also commonplace, but the intraocular gas tamponade we

used is a novel technique. Prolonged C₃F₈ gas contact with the corneal wound prevented aqueous egress, allowing sufficient wound integrity while keeping the globe formed. This is the first case report of the use of a non-expansile volume of C₃F₈ gas to repair the brittle cornea in BCS, which could be generally applied to fragile leaking corneas.

**Hussain Mohamed Hussin, Suman Biswas,
Mohamed Majid, Richard Haynes, Derek Tole**
Bristol Eye Hospital, Bristol, UK

Correspondence to: MrH M Hussin, Bristol Eye Hospital,
Lower Maudlin Street, Bristol BS1 2LX; UK;
H.M.Hussin@bristol.ac.uk

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Sympathetic ophthalmia after ruthenium plaque brachytherapy

Sympathetic ophthalmia is a rare, bilateral inflammatory process with an incidence of 0.03/100 000 in the UK and Ireland.¹ It usually follows either penetrating eye injury or intraocular surgery. Although sympathetic ophthalmia has previously been described after irradiation of ocular melanoma,^{2,3} it has never been reported after simple ¹⁰⁶Ru plaque brachytherapy. We present a case which confirms that there is a risk of developing sympathetic ophthalmia after charged-particle therapy in the absence of a penetrating injury of the uveal tract.

Case report

A 41-year-old lady was referred by her optician for a slow-growing iris lesion. Fine needle aspiration biopsy confirmed the diagnosis of ciliary body malignant melanoma. Systemic investigations for metastatic disease were negative. The patient underwent routine surgery for the insertion and subsequent removal of a ¹⁰⁶Ru plaque. Postoperative recovery was uneventful.

At 6 months after surgery, the patient presented with a 1-week history of reduced vision in both eyes to 6/24 OD and 6/36 OS. This was accompanied by photophobia, soreness and redness of both eyes.



Figure 1 The left eye after the corneal perforation was repaired. Informed consent was received for publication of this figure.