Infliximab Therapy for Aggressive Mooren Ulceration

Mooren ulceration is an uncommon corneal autoimmune disease that can lead to blindness. We describe a patient with aggressive disease in whom inflammatory control was only achieved after infusion of a tumor necrosis factor α antagonist, infliximab.

**Report of a Case.** A 37-year-old Nigerian man with a 14-year history of bilateral Mooren ulceration had a right corneal perforation and a desmetocele in his severely damaged left eye (Figure, A and B). Snellen visual acuity was 6/60 OD, improving to 6/36 OD with pinhole, and hand movements OS. Systemic cyclosporine at a dosage of 5 mg/kg/d and prednisolone tapering from 1 mg/kg/d and ceasing after 8 weeks were given, with 2% cyclosporine eyedrops and ofloxacin eyedrops both 3 times daily to the right eye. The right corneal perforation healed with glue and a therapeutic contact lens. However, the desmetocele in the left eye perforated during an inflammatory episode 2 months later, so mycophenolate mofetil (CellCept; Hoffman-La Roche Inc, Nutley, New Jersey) at a dosage of 1 g twice daily was added, a 12-mm corneoscleral lamellar tectonic graft and cataract extraction were carried out, and Baerveldt tube surgery was performed for intractably elevated intraocular pressure.

After 4 months of quiescence, the disease again reactivated with new infiltrates and melts resulting in a flat chamber in the right eye and new guttering in the periphery of the left corneoscleral graft (Figure, C and D). Cyclophosphamide treatment at a dosage of 100 mg/d was commenced with another course of oral prednisolone, and mycophenolate treatment was stopped. Three weeks later, the patient developed microscopic hematuria and bloody stools, so cyclophosphamide treatment had to be discontinued. Stool cultures were negative for parasites. The infiltrates and melts in the right eye continued to be active, so infliximab at a dosage of 5 mg/kg was added, combined with oral cyclosporine. The inflammation in both eyes responded rapidly to this treatment, with gradual return of the inflammation in the 2-week interval between infusions. The patient received 3 infusions over 6 weeks. A right conjunctival recession and cautery procedure was carried out after another perforation, which sealed spontaneously under a bandage lens, and mycophenolate was again added to the immunosuppressive regimen.

Following this, both corneas healed and the eyes became white, so the infliximab infusions were reduced to 6 weekly. At the last visit, 5 months after commencing infliximab treatment, the disease appears to be quiet (Figure, E and F). Snellen visual acuity is currently 6/60 OD, improving to 6/18 OD with a scleral contact lens. Unfortunately, the patient has no vision in the left eye due to the uncontrolled glaucoma. Infliximab infusions are currently being continued at decreasing intervals.

**Comment.** This case demonstrates how difficult Mooren ulceration can be to treat, given both the delay in onset of action and adverse effects of conventional immunosuppressive therapy. The pathogenesis of Mooren ulceration is uncertain but may involve an immune response to a corneal stromal protein (corneal antigen), which is identical to calgranulin C released by neutrophils during the host defense response to helminths. Infiltration of lymphocytes, macrophages, and Langerhans cells has been observed in the conjunctiva adjacent to Mooren ulceration.

Tumor necrosis factor α is an important cytokine in the establishment and maintenance of inflammation in many autoimmune diseases, and the success of tumor necrosis factor α antagonists in immune-mediated rheumatic diseases has led to its off-label use in ocular inflammatory disease. Apart from inhibition of tumor necrosis factor α, infliximab also appears to induce regulatory T cells in rheumatoid arthritis and to induce T-cell apoptosis in Crohn disease, both of which might be relevant in ocular inflammation.

To our knowledge, effective treatment with tumor necrosis factor α antagonists has not previously been described in Mooren ulceration. This article shows that addition of infliximab may be useful when rapid control of inflammation is desirable and/or when conventional immunosuppressive therapy is not tolerated or effective.

Valerie P. J. Saw, FRANZCO
Neil Cornelius, FCOphth(SA)
Alan D. Salama, PhD, FRCP
Charles Pusey, PhD, FRCPh
Susan L. Lightman, PhD, FRCOphth

Correspondence: Dr Saw, Moorfields Eye Hospital, 162 City Rd, London EC1V 2PD, England (v.saw@ucl.ac.uk).

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