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Rituximab treatment for peripheral ulcerative keratitis associated with Wegener's granulomatosis

Rituximab is a chimeric antibody against CD20 aimed at depleting B cells. While originally developed as a treatment for B cell lymphomas, it was recently found to be useful in managing autoimmune diseases including refractory Wegener's granulomatosis.^{1–3} We report a case of refractory peripheral ulcerative keratitis (PUK) associated with Wegener's granulomatosis that was successfully treated with rituximab.

Case report

A 57 year old man with longstanding Wegener's granulomatosis was admitted to the hospital with severe pain and blurred vision in the left eye as well as respiratory compromise because of tracheal blood clots. He had a past history of haemorrhagic cystitis while on oral cyclophosphamide and pancytopenia while on methotrexate. The diagnosis of anterior nodular scleritis and PUK was made. Treatment with intravenous methylprednisolone and monthly

Figure 1 Left eye with progressive peripheral ulcerative keratitis. An epithelial defect is present at the leading edge of the corneal thinning superotemporally. Vascular infiltration is best seen inferonasally.

Figure 2 Left eye after treatment with rituximab. The epithelial defect has healed, the corneal thinning has stabilised, and corneal vessels have started to regress.

intravenous cyclophosphamide with mesna was initiated for immediate control. While the scleritis resolved promptly, the patient had progressive peripheral corneal thinning and vascularisation. In spite of high dose oral steroids as well as anti-collagenase therapy with vitamin C and doxycycline, a chronic epithelial defect developed and the corneal thinning continued to advance (fig 1).

Because of concern over the progression of PUK and the lack of response to standard treatment, the patient received two 1000 mg infusions of rituximab separated by one week. After one week, the epithelial defect had completely resolved and the progression of corneal thinning ceased (fig 2). The eye remained stable and the patient reported significant improvement in respiratory symptoms. During a three month follow up period, the patient has been maintained free of symptoms on prednisone 10 mg daily and topical prednisolone acetate twice daily.

Comment

Rituximab is a chimeric monoclonal antibody against CD20, a marker expressed by all B cells before they mature into plasma cells. This new biological therapy acts by depleting the body's B cells and was adapted from oncology to treat autoimmune diseases. Rituximab has a good side effect profile largely limited to hypersensitivity reactions and a small increase in serious infections.^{2,4} The onset of action is as rapid as one to two weeks^{1,3} and anecdotal evidence is emerging that it can be successful in inducing lasting remission in cases refractory to other drugs. Experience in inflammatory eye disease is still sparse and we found a single case report of recalcitrant scleritis associated with Wegener's granulomatosis that responded to rituximab.⁵ In the case we describe here, the scleritis responded to high dose cyclophosphamide and steroids, but the PUK continued to progress until treatment with rituximab was initiated. While further studies are needed to determine the optimal indications and dosing for this drug, we believe that rituximab is a promising form of treatment for refractory ocular inflammation.

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Informed consent was obtained for publication of figures 1 and 2.

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NOTICES

Second Sight

Second Sight would like to hear from experienced Indian eye surgeons returning to India after training/working in the UK. Second Sight is a London based charity dedicated to the elimination of cataract blindness in India. Further details: Dr Lucy Mathen, lucymathen@yahoo.com.

2008 International Agency for the Prevention of Blindness (IAPB) 8th General Assembly

28 July–2 August, Centro de Convenções Rebouças, Sao Paulo, Brazil

Further details: Email: agency@lvpei.org.

Singapore National Eye Centre – 18th Anniversary International Meeting

14–17 March 2008, Suntec City Convention Centre, Singapore.

Further details: Tel: +65 6322 8374; Fax +65 6227 7290; Email meet@snec.com.sg.

Inaugural Asia Cornea Society Scientific Meeting

13–14 March 2008, Shangri La's Rasa Sentosa Resort, Singapore.

Further details: Fax +65 6227 7291; Email acs@snec.co.sg.

International Ocular Blood Flow Symposium

13 October 2007, Sutton Place Hotel, Toronto, Canada.

Further details: Tel: +416 978 2719 or +1 888 512 8173; Fax +416 946 7028; Email ce.med@utoronto.ca.

Neuro-Ophthalmology and Strabismus – 2008 European Professors in Ophthalmology (EUPO) Residents' Course

5–6 September 2008, Geneva, Switzerland.

This course organised by Professor Avinoam B Safran will provide an overview and an update on recent advances in neuro-ophthalmology and strabismus.

Further details: <http://eupo.eu>.