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Sir,

Anti-TNF alpha therapy in chronic necrotizing scleritis resistant to standard immunomodulatory therapy in a patient with Wegener's granulomatosis

Wegener's granulomatosis is characterized by a necrotizing granulomatous vasculitis. Ocular complications may include uveitis, scleritis, and retinal vasculitis and occur in up to 45% of patients.[1,2]

Case report

A 75-year-old man was referred to our clinic for peripheral ulcerative keratitis and anterior uveitis in both eyes. High serum proteinase-3 antibody levels in addition to a biopsy of the nasal mucosa revealing a necrotizing granulomatous vasculitis confirmed the diagnosis of Wegener's granulomatosis (WG). His visual acuity (VA) was 20/50 in the right eye (OD) and 20/100 in the left eye (OS). Systemic immunomodulatory therapy with oral cyclophosphamide (3 mg/kg/BW) with 75 mg glucocorticoids was initiated; however, despite adjustment of cyclophosphamide (4 mg/kg/BW), the scleritis remained active over the following 5 months (Figure 1a). His VA dropped to 20/200 OD and counting fingers OS. Owing to ongoing inflammation and positive reports on infliximab in systemic Wegener's granulomatosis,[3–5] infliximab was started at 5 mg/kg/BW on day 0, and repeated at weeks 2, 6, thereafter every 8 weeks. Cyclophosphamide was discontinued after week 6. After the second infliximab infusion, the anterior chamber (a/c) cell counts as well as the necrotizing scleritis decreased. At week 8, his scleritis was in remission. His VA increased to 20/60 OD and remained at counting fingers OS. Prednisolone was tapered to 12.5 mg/day (Figure 1b). After 5 months, infliximab was discontinued due to an acute herpes zoster infection, resulting in a flare-up 6 weeks later with 4+ a/c cells OD (Figure 1c) and an almost total melting of the sclera OS. His VA was counting fingers OD and light perception OS.

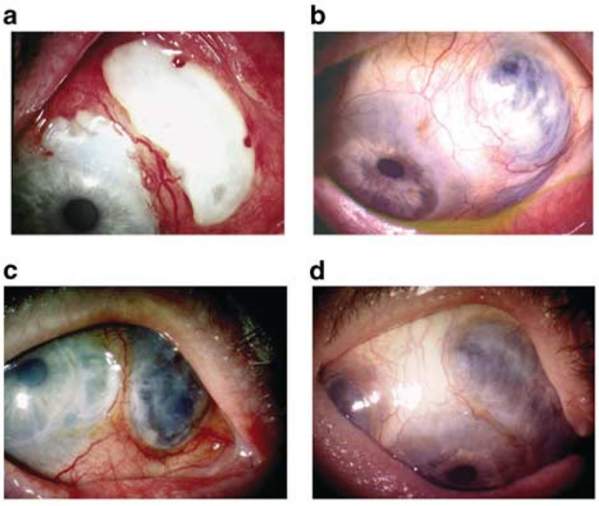

Figure 1 (a) Photograph demonstrates the active necrotizing scleritis with inflammation of the eye. (b) At 5 months after infliximab therapy has been started. The scleritis is almost completely in remission. (c) Reactivation of the scleritis after discontinuation of the scleritis. (d) At 3 months into reinstallation of combined infliximab and azathioprine therapy.

Therefore, infliximab (5 mg/kg/BW) was restarted and azathioprine at 1.5 mg/kg BW and prednisolone 50 mg/day were added. For the last 8 months, infliximab has been administered every 4–5 weeks keeping the scleritis under remission (Figure 1a). His VA increased to 20/400 in the right eye. The corticosteroid dose was tapered to 12.5 mg/day.

The classic regimen consists of cyclophosphamide and prednisone.[1,2] However, as not all patients sufficiently respond to standard therapy, and due to its toxicity alternative treatment should be considered. This is just a single case report. Still the fact that our case, in addition to previous reports, shows the effectiveness of specific TNFα-blockers in systemic WG[3–5] suggests that selective TNFα inhibition is an option in ocular manifestations of this disease.

Acknowledgements

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Sir,

Use of oral ivermectin in a patient with destructive rhino-orbital myiasis

Myiasis refers to infestation of living tissue of vertebrate animals by the eggs or larvae (maggots) of flies of the order of Diptera. Larval infestation occurs in the nose in 81% of cases, in the ear in 11%, and in the face in only 1%. Orbital myiasis cases are rare worldwide.[1–7] Conventional treatment consists of the removal of the larvae from affected sites, a painful and cumbersome task, sometimes made impossible in smaller cavities.[8,9] There have also been reports of drugs used topically to facilitate the removal, sometimes with frustrating results. We describe a patient with destructive rhino-orbital myiasis caused by larvae of the New World screwworm *Cochliomyia hominivorax* successfully treated with oral ivermectin prior to surgery.

A 55-year-old male patient was admitted to the Department of Ophthalmology, State University of Campinas with a 4-day-history of intense pain in his left orbital region, a crawling sensation, and maggots coming out of the nose. One week earlier he had mild fever, anorexia, and frontal headache. The clinical examination showed no light perception in the left eye. Discrete diffuse oedema around the nose and left orbit was noted. The eyelid was thickened and there was a necrotic, loose tissue lesion invading the left orbit (Figure 1). A purulent rhinorroea with a marked foetor was also present. Examination of the right eye was unremarkable. He was known to suffer from advanced carcinoma of ethmoidal sinus previously treated by chemotherapy and radiotherapy without success. Rhinoscopy revealed necrotic material in the nose with crawling maggots which were identified as larvae of *Cochlomyia hominivorax*. Laboratory findings were within normal limits. Computed tomography (CT) of the orbit revealed diffusely increased soft-tissue density extending to the orbital apex and obscuring orbital details. Bony destruction of the medial wall of the orbit and opacification of the ethmoidal sinus were present (Figure 2). The patient was given a single oral dose of ivermectin (200 μg/kg) and intravenous clindamicin was initiated to control associated infection. The patient showed continuous improvement and a complete resolution of the myiasis was observed after a 48-h period. Orbital exenteration was performed. No larvae were observed during the surgery or in the pathological examination of the orbital contents. The patient was discharged home on the fourth postoperative day.

Human myiasis is relatively common. It is more common in undeveloped and tropical countries, although there have been reports of myiasis all over the Planet. Most cases of myiasis probably occur when a

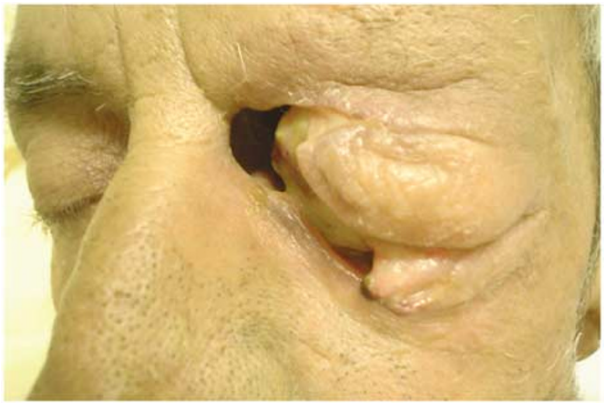

Figure 1 Appearance of lesion in the left orbit of a 55-year-old man with rhino-orbital myiasis caused by *Cochliomyia hominivorax*.