Letters to the Editor

Non-Hodgkin’s lymphoma in a patient with refractory dermatomyositis which had been treated with infliximab

SIR, The observed elevation of soluble tumour necrosis factor-alpha (TNFα) receptors in patients with dermatomyositis [1] suggests a possible therapeutic role for biological therapies directed against TNFα. We report a patient with refractory dermatomyositis, who became persistently unwell following treatment with infliximab and subsequently developed non-Hodgkin’s lymphoma.

A 48-yr-old lady presented in 1998 with arthralgia and then some months later developed a florid violaceous rash on her face and extensor aspects of the hands. Investigations at this stage demonstrated an erythrocyte sedimentation rate (ESR) of 24 mm/u/h and C-reactive protein (CRP) of < 5 mg/l. Anti-nuclear antibody was positive at a titre of 1:100 but Jo-1, Scl-70 and anticientromere antibodies were negative. Creatine kinase (CK) was normal. Skin biopsy was consistent with dermatomyositis. The skin disease failed to improve with prednisolone 30 mg daily and hydroxychloroquine 400 mg daily; subsequent treatments with methotrexate (up to 25 mg per week), azathioprine (150 mg daily) and clofazimine (100 mg daily) were similarly ineffective. Cyclosporin (5 mg/kg) was complicated by hypertension and hirsutism.

Twelve months after the initial presentation, she developed myalgia and weakness of the muscles of the shoulder girdle. At this stage she also had persisting erythema of the face, trunk and hands and peri-orbital oedema. CK was elevated at 739 U/l (normal range < 120). A percutaneous muscle biopsy did not show any features of active myositis. Intravenous pulsed cyclophosphamide produced an improvement in the muscle symptoms, with the CK returning to normal but the skin disease remained refractory. Cyclophosphamide was replaced by tacrolimus up to a dose of 2 g b.i.d. but this was withdrawn when the patient developed nausea and a sensory axonal neuropathy.

The next logical treatment was thought to be with intravenous immunoglobulin [2]. However, given recent concerns about prion transmission with human immunoglobulin [3], after discussion with the patient and informed consent, treatment was commenced with infliximab (5 mg/kg), infusions being given at 0, 2 and 6 weeks. Methotrexate 7.5 mg per week was also given. The patient, however, became increasingly unwell with anorexia and lethargy over the treatment period and developed crusting and superficial ulceration of the anterior thigh (Fig. 1). Two days after the third infusion of infliximab, she required urgent admission for further investigation. The CRP and ESR were elevated at 120 U/l and 68 mm/h, respectively (previously normal since diagnosis) and the CK was 75 U/l. Swabs of the ulceration grew methicillin-resistant Staphylococcus aureus (MRSA) and Pseudomonas aeruginosa and the appropriate antibiotic treatment was given. Infliximab was withdrawn and treatment commenced with intravenous immunoglobulin (2 g/kg). There was an improvement in the skin disease which has been sustained on monthly infusions of intravenous immunoglobulin and the ulceration has healed slowly. The patient, however, has remained systemically unwell and her CRP remains elevated (Fig. 2). Four months later, during an admission with MRSA pneumonia, she was found to have left supraclavicular lymphadenopathy and CT scan of the abdomen revealed enlarged para-aortic lymph nodes. Biopsy of the supraclavicular node has shown diffuse large B-cell non-Hodgkin’s lymphoma.

Infliximab is a chimeric human-mouse anti-TNFα monoclonal antibody that has been established in...
Concerns regarding the potential development of malignancy must be maintained. Epidemiological evidence shows an association between dermatomyositis and malignancy, including non-Hodgkin’s lymphoma [10]. Immunosuppressive therapy is also known to increase the risk of developing malignancy [11]. There have been concerns regarding the potential development of malignancy following TNF-α blockade. However, given the time-course in this patient, non-Hodgkin’s lymphoma seems more likely to be a consequence of dermatomyositis or immunosuppression but may have been unmasked by infliximab. This raises issues concerning giving infliximab to patients who have a syndrome with a recognized paraneoplastic association.

Treatment with infliximab did not produce an improvement in this patient’s skin disease. There is evidence that intravenous immunoglobulin may be useful in the treatment of refractory dermatomyositis [2] which this case supports. Infliximab was used prior to intravenous immunoglobulin in this case because of recent safety concerns regarding the potential for transmission of Creutzfeldt–Jakob disease and subsequent reduction in the availability of immunoglobulins.

We suggest that whilst evidence exists supporting anti-TNF-α therapy in certain rheumatic diseases, caution should be exercised when considering infliximab for the treatment of dermatomyositis. Further evaluation of its use in dermatomyositis is required.

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Endomyosal antibodies in psoriatic arthritis patients

Sir, We report a study of anti-endomyosal antibodies (EmA) in patients with psoriatic arthritis (PsA) in Oxford.

The presence of EmA is a useful screening test for gluten sensitivity [1], which is often unrecognized in the general population [2]. Michælsson et al. [3] found that 49 of 302 patients with psoriasis had increased serum IgA antibodies to gliadin. When 30 of these patients followed a gluten-free diet, their psoriasis improved [4]. Gluten sensitivity could be relevant to the underlying...