Clin Rheumatol (2003) 22: 369–370
DOI 10.1007/s10067-003-0744-z

LETTER TO THE EDITOR

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Infliximab in a case of early adult-onset Still's disease

Received: 8 April 2003 / Accepted: 8 April 2003

Tumour necrosis factor-α (TNF-α) blockade has recently been suggested to be effective in long-standing adult-onset Still's disease [1–4]. We have recently treated a case of early adult-onset Still's disease with the chimeric monoclonal IgG_1 antibody infliximab that we wish to describe briefly.

The patient, a 38-year-old man, was admitted to the Department of Infectious Disease in November 2002 with a 1-month history of fever, sore throat and evanescent pink maculae on his extremities that were resistant to several antibiotics. He had received follow-up examinations in the outpatient clinic of the same department for 4 years owing to inactive carriage of hepatitis B virus (HBV), with persistently normal aminotranferase levels and an IgM anti core index of < 0.1. He had had low HBV-DNA levels at PCR assay, which confers a high risk of reactivating the virus. Some days after admission arthritis involving his metatarsophalangeal joints, knees, shoulders, elbows and wrists developed. Laboratory evaluation revealed an erythrocyte sedimentation rate (ESR) of 75 mm/h, a C-reactive protein (CRP) level of 61.4 mg/dl (normal < 5 mg/dl), a serum ferritin level of 2300 ng/ml normal (< 200 ng/ml), and a white cell count of 15.4×10^9/l with 80% polymorphonuclear leukocytes. Alanine aminotransferase (ALT) was 148 U/L (normal < 40), and aspartate aminotransferase (AST) and alkaline phosphatase were normal. Results of tests for antinuclear antibodies and the rheumatoid factor were negative. The patient was given a high dose of prednisone. His condition was acceptable only after the administration of 100 mg/day. Because of the severity of the clinical situation and because HBV infection is a contraindication for methotrexate therapy, we decided to treat the patient with infliximab after obtaining his informed consent. He received the drug at a dose of 5 mg/kg by intravenous infusion at 0-, 2- and 6-week intervals and was evaluated at baseline, on days 3, 7 and 14, and then every 2 weeks. After the first infusion the patient improved and the daily steroid dosage was reduced to 50 mg. ESR, CRP and ferritin values remained elevated. After the second infusion an significant improvement was observed. The arthritis disappeared and the daily dose of prednisone was gradually tapered until it could be discontinued. ESR, CRP and ferritin values returned to normal and remained normal also after the third infusion. ALT, AST and alkaline phosphatase values did not change during the treatment. During the second infusion an urticarial rash appeared that persisted for 2 days. This appeared again during the third infusion but disappeared after a few hours. The fourth infusion will be given on an as-needed basis.

Our case confirms the efficacy of TNF-α blockage in adult-onset Still's disease [1–4]. Both main TNF-α antagonists, infliximab and etanercept, have been proved effective in the disease [1–4]. Unlike the patients of Kraetsch et al. [1], Cavagna et al. [2], and Caramaschi et al. [3] treated with infliximab, who had a long-standing disease, our patient had an early-onset disease that was resistant to steroid therapy. Similarly, the 12 adult patients with Still's disease treated by Husni et al. with etanercept had long-standing disease [4]. Therefore, future controlled studies on TNF-α blockage in adult-onset Still's disease should also include patients with early disease, because the drug might prevent the progression of articular damage. Up to 50% of patients with adult-onset Still's disease may develop a progressive and destructive polyarthritis [5, 6].

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