Sirs,

Tumor necrosis factor α (TNF-α) antagonists have been the most significant advancement in the management of Behçet’s disease (BD) (1, 2). All three available anti-TNF-α blockers have been shown to be effective on ocular (3, 4), neurological (5-7) and severe mucocutaneous manifestations (8-12). Severe, recalcitrant and therapy-resistant orogenital ulceration is a challenging problem in BD (8-10, 12). We have successfully treated a case of severe and recalcitrant orogenital ulceration by switching among the anti-TNF-α blockers.

A 47-year-old man suffering from HLA-B51 positive BD for 6 years was referred to us in February 2006 due to severe genital and ulcerous lesions which had not responded to traditional therapy including colchicine, thalidomide, cyclosporine, azathioprine and high doses of corticosteroids. Each drug was given alone or in combination at therapeutic doses and for a sufficiently long period of time. His disease was characterized by exclusive muco-cutaneous lesions including recurrent major aphthae, large and painful genital ulcerations and papulo-pustulosis.

Physical examination showed large oral aphthae involving the gengivae, the internal surfaces of the lips and cheeks and the ventral surface of the tongue, papulo-pustular lesions on the chest and large painful genital ulcerations which had not responded to us in February 2006 due to severe oral and genital malaise. The patient had difficulty in walking. Laboratory evaluation was unremarkable except for a mild increase of C-reactive protein (CRP) to 12.5 mg/L (normal <5).

After obtaining his informed consent, we began intravenous infliximab therapy at a dose of 5 mg/kg at weeks 0, 2 and 6 and every two months subsequently. After the first three infusions, the mucocutaneous lesions disappeared completely. Unfortunately the disease flared after the fifth infusion so that the sixth infusion was anticipated by a month. No results were obtained. In October 2006, the patient was given etanercept at a dose of 50 mg/week subcutaneously. The drug was ineffective also after the increase of the dose to 100 mg/week. In February 2007, we switched to adalimumab at a dose of 40 mg every other week subcutaneously. The genital ulcers and the oral aphthae were of the same severity as one year before (Fig. 1A).

There was an important improvement of the lesions after the first injection and the complete resolution after the second one (Fig. 1B). So far, the disease has remained in complete remission.

The present case permits us to make the following considerations: 1) recalcitrant and severe orogenital ulceration can successfully be treated with anti-TNF-α therapy. We confirm the observation by Robertson and Hickling (7) and Haugeberg et al. (9) who successfully treated similar cases with infliximab. 2) As suggested by Estrach et al. (8), who obtained good results on orogenital ulceration with infliximab after failing etanercept, the switch from one TNF-α blocker to another can also be useful in BD. 3) There can be a difference in the efficacy in BD between the soluble receptor etanercept and the monoclonal antibodies infliximab and adalimumab. Our patient responded temporarily to infliximab, had no response to etanercept and experienced a long-lasting remission with adalimumab. 4) Our patient was treated with anti-TNF-α therapy alone since he was resistant to conventional treatment. He had a complete remission with adalimumab. If the response had been partial we could have added azathioprin with the intention to enhance efficacy (2).

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References