Cancer complicating Behçet's disease treated with chlorambucil

Sir, O'Duffy et al recommended long term surveillance of chlorambucil treated patients with Behçet's disease for infertility, leukaemia, and other malignant lesions.1 We have seen two patients with Behçet's disease treated with chlorambucil who developed malignant disease, definite in one case and probable in the other.

The first patient, a 42 year old man, has been followed up for 14 years. The features of Behçet's disease included oropharyngeal, necrotic pseudofolliculitis, skin hypersensitivity to needle prick, and anterior and posterior uveitis with hypopyon leading to complete blindness. He was treated for five years with chlorambucil, with an initial daily dose of 0·2 mg/kg for three months and 0·1 mg/kg for 54 months. Chlorambucil was stopped because of azoospermia.

Nine years later lumbar root and right hip pains developed. Radiographs showed lytic lesions in the right ilium, the upper end of the right femur, and the pedicles of the D12 and L3 vertebrae and widening of the upper mediastinum. Fibre optic bronchoscopy was normal. There were no neoplastic cells in bronchial aspiration. Bone marrow biopsy showed infiltration with adenocarcinoma without indication of its origin. The patient died eight months later; autopsy was not performed.

Patient 2, a 44 year old man, has been followed up for three years for Behçet's disease, including oral and genital aphthosis, venous thrombosis, posterior uveitis, and hypersensitivity to needle prick. The patient was treated daily with chlorambucil in a dose of 0·2 mg/kg for 18 months, during which visual acuity and opthalmoscopic examination became normal. Nine months after cessation of treatment the patient complained of chest pains. Chest radiography showed a parahilar opacity infiltrating the pulmonary parenchyma with "cancer-leg-shaped" margins. A chest radiography had been normal nine months earlier.

Significant loss of weight was noted. Hard, irregular hepatomegaly developed, and the patient died within three months. Autopsy was not performed.

Azoospermia is frequent in chlorambucil treated Behçet's disease, occurring in 10 out of 20 patients in our experience. Leukaemia has been reported in association with cyclophosphamide treatment,2 but other cancers have not been reported previously in patients with Behçet's disease treated with immunosuppressives.

In our patients the risk of malignant change was probably increased by the administration of chlorambucil in high dose for a long time. The first patient became blind despite long term treatment with chlorambucil and developed both azoospermia and cancer. This form of treatment must be used with care and exclusively for manifestations with a poor prognosis such as severe uveitis and meningoencephalitis resistant to other treatment.

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Ann Rheum Dis 1986 45: 789
doi: 10.1136/ard.45.9.789-a

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