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Cryptococcal pleural effusion in an HTLV-I carrier with Waldenstroem's macroglobulinemia.

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Abstract

A 70-year-old woman with Waldenstroem's macroglobulinemia developed bilateral pleural effusions due to Cryptococcus neoformans. She was found to be a carrier of HTLV-I. It is speculated that the opportunistic infection occurred as the result of an impaired cellular immunity secondary to HTLV-I infection.

KEYWORDS: cryptococcal pleuritis, HTLV-1 carrier, macroglobulinemia

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— Brief Note —

Cryptococcal Pleural Effusion in an HTLV-I Carrier with Waldenstroem's Macroglobulinemia

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A 70-year-old woman with Waldenstroem's macroglobulinemia developed bilateral pleural effusions due to *Cryptococcus neoformans*. She was found to be a carrier of HTLV-I. It is speculated that the opportunistic infection occurred as the result of an impaired cellular immunity secondary to HTLV-I infection.

Key words : **Cryptococcal pleuritis, HTLV-I carrier, macroglobulinemia**

Less than 50 cases of pleural effusion due to *Cryptococcus neoformans* (CN) infection has been reported (1). We report here a case of CN pleuritis in a patient with Waldenstroem's macroglobulinemia (WMG), who had serum antibodies against human T-lymphotropic virus type I (HTLV-I), the cause of adult T-cell leukemia (ATL), and immune dysfunction, the probable predisposing factor for her cryptococcosis.

Case report

A 70-year-old female had recurrent left pleural effusion since 1986. As response was incomplete to treatment with anti-tuberculous drugs and other antimicrobics, she was referred to our clinic in May 1988. On admission, enlargement of the left cervical lymph nodes but no hepatosplenomegaly was observed. A chest radiograph showed bilateral pleural effusions (Fig 1). Serum immunoglobulin (Ig) G and A values were normal, but IgM was 27.68 g/L and monoclonal IgM, κ type gammopathy was confirmed by serum immunoelectrophoresis. Abnomal cells were not seen in the peripheral blood. A bone marrow aspirate showed 29.8 % of abnormal lymphocytes. Surface marker analysis revealed 35 % CD 19 positive cells. Biopsy of a cervical lymph node revealed diffuse infiltration of lymphoplasmacytoid cells with surface and cytoplasmic IgM, κ. A diagnosis of WMG was made. A thoracentesis revealed a reddish-yellow turbid exudate which contained predominantly normal-looking lymphocytes of helper T-cell phenotype and a IgM concentration of 13.36 g/L. Pleural fluid collected on two separate occasions grew out CN (serotype A) and contained CN antigen (titer; 1:16). None of these was detected in her serum. Indirect immunofluorescence disclosed serum antibody to HTLV-I with a titer of 1:160, but no antibody to human immunodeficiency virus. There was no delayed cutaneous hypersensitivity to purified protein derivative (PPD). Lymphocyte responses to phytohemagglutinin (PHA) and concanavalin A (Con A) were slightly

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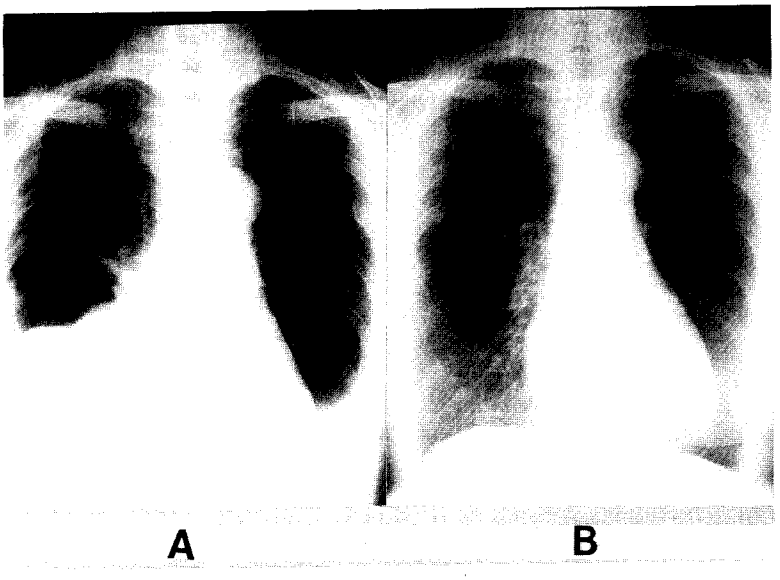

Fig. 1 Chest radiograph before (A) and after (B) treatment.

diminished (21, 287 cpm and 21,846 cpm, respectively with normal ranges of 37,700–62,400 cpm for PHA and 24,300–58,200 cpm for Con A, respectively). Intravenous (400 mg) and intrapleural (200 mg each for both sides) administrations of miconazole for three weeks failed to reduce the amount of pleural effusion, although CN could no longer be recultured and CN antigen became undetectable. Oral administration of flucytosine (1,500 mg), cyclophosphamide (100 mg) and prednisolone (15 mg) for three mosths resulted in the disappearance of pleural effusions, a decrease of serum IgM values and abnormal bone marrow cells.

Discussion

Patients with WMG may occasionally develop pleural effusion, in which malignant cells are usually seen (2). In our case, lymphocytes in the pleural fluid were mostly normal T-cells. Culture of CN and detection of CN antigen from pleural fluid suggested that CN played an etiological role of the pleuritis. Cryptococcal pleuritis is usually accompanied by primary parenchymal pulmonary disease (3). In one report of 41 cases of pulmonary cryptococcosis, 34 patients were compromised hosts (4). In multiple myeloma, a disorder related to WMG, cellular immunity is normal and viral or fungal infections are rare (5). Nevertheless, our patient had impaired cellular immunity, as judged by a negative PPD skin test and suppressed lymphocyte responses to mitogens. It is likely that her impaired cellular immunity arose from HTLV-I infection of T4 lymphocytes. This assumption may be supported by reports of *Pneumocystis carinii* pneumonia (6), a high *Strongyloides stercoralis* infestation rate (7), an increased risk of malignancy (8) and dissemination of early cancer in HTLV-I seropositive persons (9). In regions where HTLV-I infection is prevalent, serological testing for HTLV-I is recommended in persons developing opportunistic infections.

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