Cryptococcal Meningitis in a Patient with Chronic Adult T-cell Leukemia in Complete Remission

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Introduction

Adult T-cell leukemia (ATL) is a neoplastic disease caused by human T-lymphotropic virus type I (HTLV-I) infection1). Although infectious complications are known to occur in patients with ATL, infections in ATL remission are poorly documented. We report an autopsied patient with chronic ATL in continuous complete remission who died of cryptococcal meningitis.

Case Report

A 73-year-old woman was admitted to our hospital with a loss of consciousness and 38°C fever on April 22, 1997. Chronic ATL was first diagnosed in 1990, when intermittent oral administration of cyclophosphamide and prednisolone was started. Complete remission was confirmed in physical and hematological examinations several months later, and this was maintained without chemotherapeutic agent. However, HTLV-I antibody remained positive, as did HTLV-I proviral DNA. On admission, she had an increased leukocyte count at 15 \times 10^9/l and serum CRP at 7.57 mg/dl. Infectious meningitis was diagnosed from neck stiffness and increased pressure (25 cmH2O), cell count (polynuclear 415/3 and mononuclear 96/3), and protein (350 mg/dl) in the cerebrospinal fluid (CSF). Daily intravenous administration of 6 g of piperacilline, 2 g of panipenem/betamiprone, 200 mg of fluconazole, and 600 ml of 10% glycerol provided no

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Fig. 1 Cryptococcal bodies seen in the meninx (PAS stain, ×400).
benefit and the woman died of sudden respiratory arrest on the hospital day 12. After written permission for autopsy, we found brain stem herniation to have directly caused her death. The pia and arachnoid were cloudy and thickened macroscopically. Histological examination showed cryptococcal dissemination to the meninx (Fig. 1) and the lung, although the culture of venous blood and CSF on admission was negative. No ATL cells were detectable in any tissues.

**Discussion**

HTLV-I-infected lymphocytes are reported to be functionally deficient\(^2\)\(^3\), and opportunistic infections due to fungus or pneumocystis carinii are observed even in subclinical HTLV-I carriers\(^4\)\(^5\). These studies indicate that patients in ATL remission are still immunocompromised. Kouno et al. reported that 6 of 19 (32%) patients with pulmonary cryptococcosis were positive for HTLV-I antibody\(^6\). Taken together, those with HTLV-I, including carriers and remitted ATL patients, who present meningitis symptoms should be empirically and intensively treated with antifungal agents, even if the CSF culture is negative. We also recommend that HTLV-I antibody must be checked for in fungal meningitis despite the absence of disease known to generate immunodeficiency.

**References**

慢性型成人 T 細胞性白血病の完全覚解期に生じたクリプトコッカス菌膜炎の一例

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成人 T 細胞性白血病（ATL）における種々の日和見感染合併が知られているが、覚解期の易感染性について検討した報告は少ない。我々は慢性型 ATL の完全覚解例に併発したクリプトコッカス菌膜炎を経験した。症例は 73 歳女性、1997 年 4 月 22 日、意識消失と発熱のため当科入院、頸部硬直、髄液細胞数増加などから菌膜炎として抗生剤、抗真菌剤を投与したが、第 12 病日に死亡、剖検で菌膜、肺にクリプトコッカス菌が証明された。HTLV-I キャリアにも真菌感染症を生じることが報告されており、HTLV-I 陽性者が菌膜炎症状を呈した際にはキャリアや覚解例であっても速やかに強力な抗真菌治療を行うべきであると考えられた。

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