中文題目：一個少見的雙側肺結節案例

英文題目：An unusual case of multiple lung nodules

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Background

Lymphomatoid granulomatosis is a form of lymphoproliferative disorder associated with Epstein-Barr virus (EBV) infection in most cases. Lung is the most commonly involved organ. We presented a case of pulmonary lymphomatoid granulomatosis in a previous healthy man.

Case presentation

A 53-year-old male patient without any systemic disease before presented with general malaise and body weight loss with 11 kilograms (13%) in 3 months. Left pleural effusion with bilateral multiple lung nodules were noticed on chest X-ray during health examination. Intermittent low-grade fever developed after that. Chest computed tomography revealed multiple roundish nodules about 2-3 centimeters in diameter in both lungs with lower lung field predominant. Infectious disease, primary lung malignancy, or metastatic lesion was considered at beginning. Ampicillin/sulbactam was prescribed empirically but his fever persisted. Pleural effusion analysis showed exudative effusion with mononuclear cells predominant (lymphocyte 37%, monocyte 30%). Computed tomography-guided tumor biopsy, and bronchoscopy with transbronchial biopsy were performed but the pathology report showed only inflammatory and necrotic tissue found. Owing to no definite diagnosis, video-assisted thoracoscopic wedge resection of left upper and left lower lung was arranged. The pathology report revealed lymphomatoid granulomatosis, grade 3, which was confirmed by Epstein–Barr encoding region (EBER) in situ hybridization. He received chemotherapy with R-CHOP (rituximab, cyclophosphamide, epirubicin, vincristine, and prednisolone) thereafter. The bilateral lung nodules and left side pleural effusion got improved after systemic chemotherapy.

Discussion

Lymphomatoid granulomatosis is an angiodestructive EBV-positive lymphoproliferative disease. It primarily affects the pulmonary system with nodular lesion over bilateral lung fields along the bronchovascular structures or interlobular septa with lower lung predominance distribution. The pathology would exhibit atypical EBV-positive B cells with other mononuclear cells comprised of T cells and plasma cells in the background. Lymphomatoid granulomatosis is divided in three grades according to the proportion of EBV-positive B lymphocytes by EBER in situ hybridization.

Our case is a male patient with lymphomatoid granulomatosis categorized as grade 3 (more than 50 EBV-positive cells per high power field). It is hard to diagnose without adequate tissue that our case finally receives video-assisted thoracoscopic lung wedge resection. It’s a rare disease and difficulty in confirming diagnosis. Physician should keep in mind the clinical and radiological feature of lymphomatoid granulomatosis when multiple lung nodules were noted in the chest X-ray or chest computer tomography. Discussion with the pathologist is also important to make the correct diagnosis.