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CASE REPORT

An unusual cause of pulmonary nodules: Pulmonary hyalinizing granuloma with recurrence

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Summary
We present a case of pulmonary hyalinizing granuloma (PHG) associated with a history of pleural effusion, autoimmune thyroiditis and spontaneous ecchymosis especially periorbitally. She presented with complaints of dyspnea, cough and chest pain. Two right-sided pulmonary nodules were resected and proved to be PHG histopathologically. After a 2 years follow up, we have seen multiple bilateral nodules of varying size which were considered as recurrence by excluding all possible primary or metastatic malignancy.

Introduction

Pulmonary hyalinizing granuloma (PHG) is a rare disease characterized by bilateral or unilateral nodules, sometimes with cavitation and/or calcification which was first described in 1977 by Engleman et al.[1] Although the exact etiology is unknown, it is proposed that this disorder is a result of exaggerated reaction to antigens such as histoplasma, mycobacterium or aspergillus.[1–3] Less than 100 cases have been published and only a few of them reported recurrence.[4]

Case report

A 37 years old woman was referred to our department with abnormalities on chest roentgenogram, pulmonary and nonpulmonary symptoms. She had a very extensive medical history: spontaneous ecchymosis (especially periorbital) for last 4 years, right exudative pleural effusion with a nondiagnostic closed pleural biopsy 3 years ago, hypothyroidism, autoimmune thyroiditis with anti-thyroglobulin and anti-microsomal antibody (AMA) positivity. She was taking

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L-tyroxine. Her chief complaints were right-sided chest pain, nonproductive cough and hemoptysis when she was admitted to our hospital in June 2003. On physical examination, body temperature was 36.5 °C, blood pressure was 110/60 mmHg, heart rate was 72/min and respiratory rate was 22/min. There was ecchymosis on periorbital region and no other abnormalities on systemic examination. Laboratory evaluation disclosed an erythrocyte sedimentation rate of 28 mm/h, normal hemoglobin, hematocrit and white blood count. Blood chemistry including thyroid, liver and kidney function tests were within normal limits. Antinuclear antibody (ANA), anti-dsDNA antibody, anti-smooth muscle antibody (ASMA) rheumatoid factor and anti-cytoplasmic antibodies (*c*- and *p*-ANCA) were negative. Tumor markers were also negative. Peripheral blood smear was normal and Coombs' test was negative. Purified protein derivative (PPD) skin test was positive producing 20 mm induration while acid-fast bacillus and culture for tuberculosis were negative in sputum.

Posteroanterior and right-sided chest X-rays revealed two cavitating nodules with ill-defined margins. Computed tomography (CT) of the chest showed the nodules in the subpleural location (Fig. 1). According to earlier tomography, the nodules were smaller without any cavitation in 2000. Fiberoptic bronchoscopy showed no endobronchial lesion and tru-cut biopsy was done to the upper lobe nodule. The cytopathologic examination revealed ''acellular necrotic material with hyalinization''. Then, right upper lobe nodule was excised with wedge-resection. Macroscopic examination of the excised nodule showed a firm fibrous capsule and cavity with necrotic material. Microscopically, the lung tissue was replaced with hyalinizing acellular haphazard collagen bundles among which necrosis and dystrophic calcification were present. There were interstitial fibrosis, mononuclear cellular infiltration and vascular wall thickening in the surrounding lung tissue: PHG (Fig. 2). Following this diagnostic operation, 1 month later, the nodule in the lower lobe was excised by wedge-resection because surgery was the only therapeutic option and the pathologic diagnosis was the same.

After 2 years, cough and weakness had resolved but chest pain persisted and hemoptysis recurred. CT of the chest on June 2005 revealed multiple, bilateral nodules of varying size and some of nodules were cavitating (Fig. 3). To rule out malignancy, a full work-up that consisted physical examination including abdominal, pelvic, breast and head and neck exams, ultrasonography of abdomen, thyroid, pelvis and

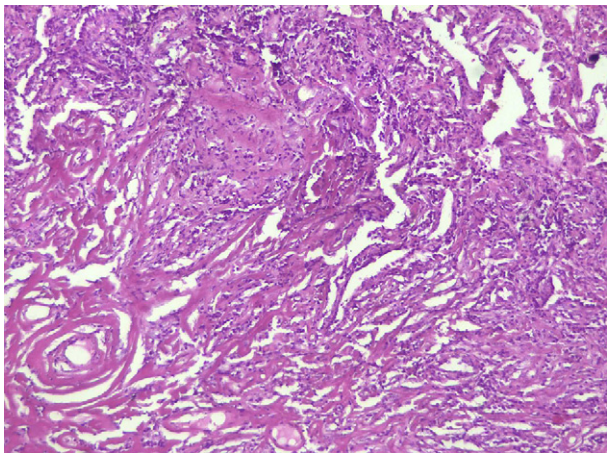

Figure 2 Microscophic appearance of resected right upper lobe nodule: Hyalinized lamellae surrounded by inflammatory cell deposition (*H–E* × 100).

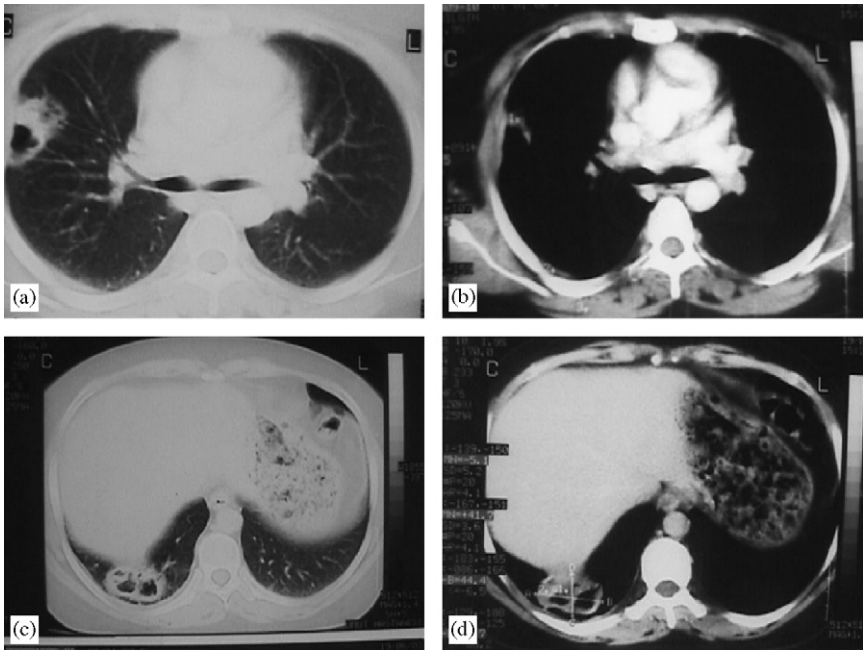

Figure 1 Computed tomography of chest showing cavitating subpleural nodules in right upper (a and b) and lower (c and d) lobes with some perinodular infiltration.