Inflammatory Pseudotumor of the Lung with Rapid Growth

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Abstract

A 58-year-old man presented with a nodule in the right lung. Initially, the chest CT showed a ground-glass shadow. However, the shadow had become a solid nodule one month later. Histological examination revealed it was an inflammatory pseudotumor of the lung and subsequent surgery showed it to be an organizing pneumonia type. This disease is rare and in most cases is considered to be slow growing. Chest CT findings in the early stages have not been reported previously. Therefore, the present case is noteworthy in that we could confirm the CT findings in the early stages of this disease with rapid growth.

Key words: chest CT findings, early stages, organizing pneumonia type


Introduction

Inflammatory pseudotumor of the lung is a rare disease. The disease is benign and most commonly presents as a localized and solitary nodule. This non-malignant lesion consists of mixed components, comprising collagen fibers, and inflammatory and mesenchymal cells. In most cases, inflammatory pseudotumors of the lung are slow growing (1). To our knowledge, no cases of rapid growth, with chest CT findings in the early stages of this disease, have been reported previously. We report here the early chest CT findings of a case of inflammatory pseudotumor of the lung with rapid growth. The histopathological findings of the present case are interesting considering the likely pathological progression of this disease.

Case Report

A 58-year-old man had a medical checkup and his chest CT scan revealed a small area of ground-glass opacity in the S4 region of the right lung. He was reexamined one month later when the chest CT scan revealed that the lesion with ground-glass opacity had transformed into a 15 mm-diameter solid nodule. This nodule was suspected to be a malignant lesion. A physical examination identified no abnormalities that suggested the presence of pneumonia. Laboratory data, including tumor markers, were within normal limits. White blood cell count was 5,800/mm3 with 59.3% neutrophils and C-reactive protein was 0.1 mg/dL. These data were considered to indicate no existence of inflammation. 2-[18F] fluoro-2-deoxy-D-glucose and positron emission tomography (FDG-PET) showed a weak accumulation of FDG indicating the possibility of a malignant lesion. SUVmax of this lesion in FDG-PET was 3.2. However, a transbronchial biopsy, which was performed twice, failed to confirm the diagnosis. A further CT scan was then performed which showed rapid enlargement of a lesion (Fig. 1). This rapid enlargement was still considered as possibly malignant, and therefore an open lung biopsy by right middle lobectomy was performed. Macroscopic findings showed a tumor, 20 mm in diameter, with a whitish appearance and well-defined edges. Histological examination revealed that the nodule consisted of a diffuse infiltration of inflammatory cells, lymphocytes, histiocytes and undifferentiated fibroblasts. Granulomatous changes were seen in the alveolar areas. No cytological dysplasia was found and no apparent alveolar destruction was observed (Figs. 2A, 2B). As a result, the tumor was diagnosed as an inflammatory pseudotumor of the lung, which is an organizing pneumonia type. The patient
Figure 1. The time course of the chest CT scan shows rapid growth of the lesion. The very early stages of the CT findings indicate ground-glass opacity.

Figure 2. Histological findings reveal the diffuse infiltration of inflammatory cells, lymphocytes, histiocytes and undifferentiated fibroblasts. Granulomatous changes occurred, but no cytological dysplasia or apparent alveolar destruction was observed. The features led to the diagnosis of inflammatory pseudotumor of the lung, organizing pneumonia type. Hematoxylin and Eosin staining, A) Low magnification. B) High magnification.

was discharged from our hospital without any post-operative complications.

Discussion

The incidence of inflammatory pseudotumor of the lung is considered to be extremely rare. For example, in a survey of 56,400 thoracic surgery cases, only 0.04% of patients had this diagnosis (2). The inflammatory pseudotumor is generally characterized by a localized and solitary nodule. This nonneoplastic lesion consists of mixed components, including collagen fibers, and inflammatory and mesenchymal cells. In 1988, Matsubara et al (1) proposed that inflammatory pseudotumor of the lung could be divided into three subtypes according to the major histopathologic features: organizing pneumonia type, fibrous histiocytoma type and lymphoplasmacytic type. In brief, organizing pneumonia type is characterized by intraalveolar lymphohistiocytic inflammation, which converts to intraalveolar fibrosis peripherally, and interstitial fibrosis centrally, because of a proliferation of fibroblasts. The fibrous histiocytoma type is characterized by a predominant proliferation of spindle cells and histiocytes showing a storiform pattern, with a loss of alveolar architecture. The lymphoplasmacytic type is characterized by a predominance of lymphocytes and plasma cells with little fibrosis. They also postulated that this disease might originate as the organizing pneumonia type and subsequently transform to either the fibrous histiocytoma type or lymphoplasmacytic type (1).

Matsubara et al (1) also described that the inflammatory pseudotumor of the lung grew slowly; however, there have been few reports that have discussed the pathological course of this disease. Furthermore, to our knowledge, there have been no case reports of this disease that have shown rapid
growth. Some reports have described the radiologic features of inflammatory pseudotumors of the lung. For example, Agrons et al (3) investigated 61 cases of this disease and Kim et al (4) discussed 10 cases; however, in neither report were any early chest CT findings presented. Therefore, the early radiographic features, and hence the early pathological features, of this disease have not yet been clarified.

We summarized 7 cases of inflammatory pseudotumor of the lung that have been reported over the last decade where we could confirm the time course of the disease (Table 1). In 4 cases, slow growth of the lesions was observed over several years (5-8). In contrast, in the other 3 cases growth was observed in under a year (3, 9, 10). For all 3 cases, the masses had already grown to some extent at the first finding and therefore the opportunity to investigate the early lesions by chest CT scan had passed.

Here, we have presented a case of inflammatory pseudotumor of the lung with rapid growth. We consider the present case lends support to the hypothesis of Matsubara et al that inflammatory pseudotumor of the lung might originate as the organizing pneumonia type. We cannot confirm the correctness of Matsubara’s hypothesis based on the experience of only one case. Nevertheless, the course of the present case is noteworthy because it represents the very early stage of inflammatory pseudotumor of the lung, indicative of the organizing pneumonia type, and this finding is unprecedented. Little is known about the precise mechanisms of rapid growth in the present case. Some part of the mechanisms might depend on the cell type. The cells in “organizing pneumonia type” phase might grow rapidly and then transform to the cells in the “fibrous histiocytoma type” or “lymphoplasmacytic type”. The cells in this phase might stop growing fast.

As discussed above, there have been no case reports of this disease with rapid growth that also presented the early radiologic findings of this disease. Through a study of the present case using chest CT scans, we were able to observe the very early stages of an inflammatory pseudotumor of the lung. The scans were able to demonstrate the rapid growth from a faint ground-glass shadow to a solid nodule. The doubling time of the lesion was only 7 days. In one case report describing an inflammatory pseudotumor, the observation was made that a doubling time of between 20 and 400 days suggested a malignant lesion, whereas a doubling time of less than 20 days suggested a benign one (9). Taking this observation into consideration, the short doubling time of the present case pointed to the likelihood of a benign tumor before the operation confirmed it.

In summary, we observed the very early stages of an inflammatory pseudotumor of the lung with rapid growth both radiographically and histologically, under circumstances of which there has been no previous experience.

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