We report a relatively rare surgical treatment for two cases of inflammatory pseudotumors of the lung. In case 1, a 52-year-old male with a history of left chest pain was admitted to our hospital for an abnormal nodule with an irregular margin that was detected in the left upper lung field. The nodule, measuring 15 mm in diameter, was larger than the one observed six months earlier, which had been removed by a thoracoscopic resection. In case 2, a 64-year-old female with a history of chronic cough and hemoptysis was admitted to our hospital, and an abnormal nodule with pleural indentation was detected in the lower left lung field. The nodule, measuring 8 mm in diameter, was also removed by a thoracoscopic resection. In both cases, the histologic examination enabled us to diagnose the lesion as an inflammatory pseudotumor. In general, it is very difficult to differentiate inflammatory pseudotumors from malignant tumors of the lung. The best treatment for inflammatory pseudotumors is usually early and complete surgical resection, since it can lead to improved survival. Therefore, we consider thoracoscopy-aided surgery to be less invasive and more useful than other surgical methods in the diagnosis and treatment of inflammatory pseudotumor of the lung.

Key words: inflammatory pseudotumors, thoracoscopic resection

Introduction

Inflammatory pseudotumor of the lung is a relatively rare benign tumor of unknown origin, which occasionally presents as an aggressive, proliferative tumor that may begin as organizing pneumonia. We report two cases treated by thoracoscopic resection, in which it was very difficult to differentiate inflammatory pseudotumors from malignant tumors of the lung. These cases are discussed along with a review of other cases in the Japanese literature that were diagnosed by thoracoscopic resection.

Case Report

Patient 1.

A 52-year-old male with a 1-month history of left chest pain was diagnosed with pleurisy, who was given 20 mg of prednisone to be taken daily. During a 1-month follow-up examination, we found an abnormal nodule with irregular margin in the left upper lung field, and the patient was admitted to our hospital. Six months earlier, a smaller nodule measuring 15 mm in diameter had been detected on a chest radiograph and computed tomography (CT) scan (Fig. 1). Laboratory examinations at admission, including a test for identifying serum tumor
markers did not show any abnormalities. Because the pathological diagnosis was not established, and lung cancer was suspected preoperatively, based on the radiographic findings, we performed a thoracoscopic resection. The histologic examination demonstrated spindle-shaped fibroblasts and myofibroblasts, arranged monotonously without mitosis or necrosis, including infiltrations of plasma cells, which confirmed the diagnosis of an inflammatory pseudotumor (Fig. 2). Clinical characteristics were uneventful, and the patient was discharged. Five years later, he remains well without signs of tumor recurrence.

**Patient 2.**

A 64-year-old female with a 10-day history of chronic cough and hemoptysis was admitted to our hospital and an abnormal nodule, measuring 8 mm in diameter, with pleural indentation was detected in the left lower lung field on a chest radiograph and CT scan. The nodule measured 8 mm in diameter (Fig. 3). Laboratory examinations performed on admission, including those for identifying serum tumor markers, showed no abnormalities. The tumor was rated as class II by Bronchofiberscopic cytology was done, which was reported as a class III. But the patient underwent thoracoscopic resection because of suspected lung cancer observed in the CT scan image and positron emission tomography (PET) with F-18 fluorodeoxyglucose (FDG), in which the SUVmax was 2.70 (Fig. 4). The histologic examination diagnosed the growth as an inflammatory pseudotumor, which included spindle-shaped fibroblasts and myofibroblasts, heavy infiltrations of lymphocytes, plasma cells, and histiocytes (Fig. 5). The patient had uneventful sequelae and was discharged. There was no recurrence during one year of follow-up.

**Discussion**

Inflammatory pseudotumor is a relatively rare benign tumor of the lung with an unknown etiology, accounting for 0.7% of primary pulmonary and bronchial tumors. Controversy about the histogenesis of inflammatory pseudotumor has produced a variety of synonyms, including plasma cell granuloma, plasma cell tumor, inflammatory myofibroblastic tumor, histiocytoma, xanthoma, and fibrous xanthoma. Although this is a benign tumor of unknown origin, with no predisposition to gender or race, the lesions are often locally invasive, requiring extensive pulmonary resection to prevent local recurrence, considerable biologic aggressiveness and unfavorable evolution. Death is secondary to both local relapse with infiltration of the mediastinal organs and distant metastases.

After reviewing the Japanese literature for surgical cases of inflammatory pseudotumor of the lung treated by thoracoscopic resection since 1998, we could only find
11 cases, including our two cases (Table 1). As shown in Table 1, the mean age of the patients was 49 ± 13 years, with an age range of 26 to 65 years; four patients were male and seven were female. The mean maximum tumor size was 2.0 ± 0.8 cm, with a range of 0.8 to 3.5 cm. The initial consultation in seven patients (64%) was admission with respiratory complaints, such as a cough in two cases, hemoptysis in two cases, chest pain in two cases, and fever in two cases. Lesions were detected on medical check-up in three patients (27%) and on follow-up for other disease in one patient (9%). Symptomatic incidence in our review of surgical cases was higher than that in other series, in which the discovery was incidental on examination of radiographs obtained for other purposes.

Cefolio et al. reported that the tumors could be divided into the two types, based on the presence or absence of local invasion of vessels. The first type is the noninvasive type, which is asymptomatic and the second type is the invasive type, which is symptomatic. Symptoms seem to be strictly related to lesion location: parenchymal, mediastinal, or endobronchial. We consider that it might be more likely for a patient to undergo thorascoscopic resection for a diagnosis of disease rather than for relief of symptoms.

It had been suggested that radiologically nodules typically present as solitary circumscribed masses with a round or oval shape. However, lung cancer is often suspected preoperatively, based on the radiographic findings. Indications for thorascoscopic resection to rule out malig-

Fig. 3 Computed tomography (CT) scan showed an abnormal nodule with pleural indentation in the left lower lung field on chest radiograph and measuring 8 mm in diameter.

Fig. 4 An abnormal nodule with pleural indentation in the left lower lung field was suspected of bronchogenic carcinoma on positron emission tomography (PET) with F-18 fluorodeoxyglucose (FDG) imaging in which a SUVmax was 2.70.

Fig. 5 Histologic examination showed that this tumor was composed of spindle-shaped fibroblasts and myofibroblasts with heavy infiltrations of lymphocytes, plasma cells, and histiocytes and was diagnosed as an inflammatory pseudotumor (H-E stain x 400).
nancy is suggested by chest CT scanning in all patients, such as a nodule with irregular shape in four cases (36%), nodule with spicula in two cases (18%), nodule with pleural indentation in two cases (18%), nodule only in two case (18%) and nodule with cavity in one case (10%). The computed tomographic scan did not facilitate identification of any specific features of inflammatory pseudotumors of the lung, and the patients underwent surgery for suspected lung cancer or carcinoid from a tentative diagnosis. In both of four cases, the method of diagnosis was a thoracoscopic resection.

Our surgical procedure was a partial resection (wedge resection) for 10 cases and lobectomy for one case. During the long-term followup, there was no recurrence of growth after the partial resection with complete removal of the inflammatory pseudotumor of the lung (Table 1). Partial resection, if radical, is a suitable curative procedure. The best treatment for inflammatory pseudotumors should be early and complete surgical resection, because these lesions are often locally invasive and complete surgical resection can lead to an improved prognosis and survival. The noninvasive type is characterized by a small size that does not invade surrounding structures and is usually removed by partial resection. The invasive type is usually large size and may invade local mediastinal structures or chest wall and may require anatomic pulmonary resection such as segmentectomy or lobectomy to avoid local recurrence.

In particular, when inflammatory change around the bronchus is remarkable, thoracotomy may be performed for complete removal of the tumor.

Recently, developments in video camera technology along with advances in percutaneous endoscopic instruments have expanded the role of diagnostic video-assisted thoracic surgery (VATS) for such small lung nodules. It has been reported that VATS lung biopsy offered virtually 100% sensitivity and specificity, and avoided thoracotomy for benign disease. However, it has also been reported that PET with FDG provides useful information on malignant lesions. However, a recent study showed that FDG PET had a low specificity since FDG uptake can be observed at sites of active, acute inflam-

<table>
<thead>
<tr>
<th>Author (year)</th>
<th>Age / Sex</th>
<th>Chief complaint</th>
<th>Location</th>
<th>Size</th>
<th>CT features</th>
<th>surgical procedure</th>
<th>Recurrence (months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1) Nakashima (1998)</td>
<td>27 / F</td>
<td>chest abnormal shadow</td>
<td>LLL (S8 &amp;S9)</td>
<td>S8: 2cm, S9: 1 cm</td>
<td>nodule with pleural indentation</td>
<td>partial</td>
<td>-</td>
</tr>
<tr>
<td>2) Fukuhara (1998)</td>
<td>56 / F</td>
<td>chest abnormal shadow</td>
<td>RML (S5)</td>
<td>1.2 x 1.0 cm</td>
<td>nodule with spicula</td>
<td>partial</td>
<td>-</td>
</tr>
<tr>
<td>3) Watanabe (1999)</td>
<td>26 / F</td>
<td>chest abnormal shadow</td>
<td>RML (S4)</td>
<td>2.5 cm</td>
<td>nodule</td>
<td>lobectomy</td>
<td>none (21)</td>
</tr>
<tr>
<td>4) Udaka (1999)</td>
<td>52 / M</td>
<td>fever</td>
<td>LUL (S1 + 2)</td>
<td>3.0 x 2.0 cm</td>
<td>nodule with a cavity</td>
<td>partial</td>
<td>none (21)</td>
</tr>
<tr>
<td>5) Hanaoka (1999)</td>
<td>56 / F</td>
<td>hemoptysis</td>
<td>RLL (S9)</td>
<td>1.5 cm</td>
<td>nodule with pleural indentation</td>
<td>partial</td>
<td>none (26)</td>
</tr>
<tr>
<td>6) Hanaoka (1999)</td>
<td>49 / M</td>
<td>chest abnormal shadow</td>
<td>LUL (S4) &amp; LLL (S8)</td>
<td>S4: 1.4 x 1.1 cm, S8: 2.7 x 2.2 cm</td>
<td>nodule with irregular margin</td>
<td>partial</td>
<td>none (9)</td>
</tr>
<tr>
<td>7) Araki (2003)</td>
<td>65 / F</td>
<td>cough</td>
<td>RUL (S3)</td>
<td>1.7 x 1.5 cm</td>
<td>nodule with pleural indentation</td>
<td>partial</td>
<td>none (11)</td>
</tr>
<tr>
<td>8) Mizuno (2006)</td>
<td>53 / M</td>
<td>fever, cough</td>
<td>RLL (S6)</td>
<td>3.5 x 2.0 cm</td>
<td>nodule with irregular margin</td>
<td>partial</td>
<td>-</td>
</tr>
<tr>
<td>9) Nakamura (2007)</td>
<td>38 / F</td>
<td>anterior chest pain</td>
<td>RML</td>
<td>2.8 cm</td>
<td>nodule</td>
<td>partial</td>
<td>-</td>
</tr>
<tr>
<td>10) Author (2009)</td>
<td>52 / M</td>
<td>chest pain</td>
<td>LUL (S1 + 2)</td>
<td>1.8 cm</td>
<td>nodule with irregular margin</td>
<td>partial</td>
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<tr>
<td>11) Author (2009)</td>
<td>64 / F</td>
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<td>LLL (S8)</td>
<td>0.8 cm</td>
<td>nodule with pleural indentation</td>
<td>partial</td>
<td>none (25)</td>
</tr>
</tbody>
</table>

CT, computed tomography; LLL, left lower lobe; RML, right middle lobe; LUL, left upper lobe; RUL, right upper lobe; RLL, right lower lobe
formation. Therefore, it can not offer the virtually 100% sensitivity and specificity offered by VATS as shown in case 2.

Therefore, we consider thoracoscopy-aided surgery, which is less invasive, to be the most useful procedure for early diagnosis and treatment of inflammatory pseudotumor of the lung.

References