INFLAMMATORY PSEUDOTUMOR OF THE LUNG:
REPORT OF FOUR ADULT CASES

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Inflammatory pseudotumors of the lung are rare and may produce diagnostic and therapeutic dilemma for the clinicians, pathologists, and surgeons. Herein, we reported on four patients with this uncommon disease. They were three men and one woman with a mean age of 37 years (range: 20 – 50). Three cases were symptomatic, complaining of cough, expectoration of sputum, hemoptysis, and chest pain. Three had solitary pulmonary nodule (SPN) and one had a tumor-like mass in chest X-ray films. Wedge resection was performed for the first three cases. With suspicion of lung cancer, pneumonectomy was carried out for the last patient.

The overall survival was 100%. There was no operative or postoperative complications/death or recurrence within 5 – 10 years of follow-up. All of them had benign appearances, which was proven by pathologic examination.

Inflammatory pseudotumors of the lung are rare. Most behave in a benign fashion. It may increase slowly in size and sometimes has an aggressive behavior. Frozen section may be used during the operation for the diagnosis. According to pathologic examination, complete resection is safe and leads to excellent survival.

Keywords: Inflammatory pseudotumor • lung • plasma cell granuloma • solitary pulmonary nodule (SPN)

Introduction

Inflammatory pseudotumors of the lung are rare benign tumors which are in fact, non-neoplastic unregulated growth of inflammatory cells. Occasionally, aggressive forms are seen.1, 2, 3 Umiker and Iverson recognized this entity for the first time and named it “postinflammatory tumors of the lung”.3, 4 The lung and airways are involved in the majority of cases. Mediastinum, thoracic lymph nodes, and other structures are rarely affected. As a localized tumor, benign proliferation of predominantly plasma cells with reticuloendothelial elements is often seen, so it is sometimes called “plasma cell granuloma”.5 – 7

However, as Colby et al have pointed out, this term is better reserved for one of the two major types of inflammatory pseudotumor of the lung. The other major type is called fibrohistiocytic.9 Inflammatory pseudotumors have been also called histiocyteoma, xanthoma, xanthogranuloma, fibroxanthoma, mast cell granuloma, and pseudolymphoma because of proliferation of other cell types.2, 4, 5, 9, 10

Inflammatory pseudotumors may mimic lung carcinoma and pose diagnostic and therapeutic difficulties.6, 9 These are the most common primary lung tumors in children and should be kept in mind as the differential diagnosis of every SPN or lung mass.2, 5

Case Report

Four patients with inflammatory pseudotumor of the lung were operated upon in the Thoracic Surgery Department of Imam Khomeini Hospital, Tabriz, Iran, between 1980 and 1990. There were three men and one woman with a mean age of 37...
years (range: 20 – 50). Three were symptomatic, complaining of cough, expectoration of sputum, hemoptysis, and chest pain. The other one was asymptomatic. On physical examination, only one patient had clubbing and decreased breath sounds on the affected side. Bronchoscopy was negative in all four cases. Cytologic examination of the sputum and bronchoalveolar lavage fluid were negative for malignant cells. Routine hematological and biochemical tests as well as urinalysis were unremarkable. Three patients had a solitary pulmonary nodule (Figure 1); one had a large mass with undefined margins, resembling lung cancer on chest roentgenogram (Figure 2). Wedge resection was performed for the first three cases; pneumonectomy was carried out for the last one who had a gross appearance of malignancy. Pathological study of these specimens revealed a benign inflammatory pseudotumor of the lung (Figures 3 and 4). All patients were followed for 5 – 10 years. There was no operative or postoperative death. No recurrence or major complications were noted in the follow-up examination.

**Discussion**

It is difficult to obtain a true incidence of pulmonary inflammatory pseudotumors. Golbert and Pletnov reported an incidence of 0.7% in their study on 1,075 pulmonary and bronchial tumors conducted in Hertzen Moscow Oncological
Institute. A much lower incidence of 0.04% is reported in some other series. It has been seen in all age groups, but are the most common primary tumor-like lesions of the lung in children aged less than sixteen years. Our patients aged between 20 and 50 years. The etiology of the disease still remains obscure. Three of four patients (75%) were symptomatic. A lower incidence of symptomatic patients, however, was reported in larger series. The heart, stomach, breast, and pleura are sometimes reported to be involved. Fever and clubbing have been reported and generally disappear after resection of the lesion. Bronchoscopy and cytological examinations of the sputum are often normal. Most pseudotumors are seen in the periphery of the lungs as SPN or a mass. Sometimes, calcification, cavity formation, and hilar lymphadenopathy may be seen. Pleural effusions—typically small and ipsilateral—could be found in up to 13% of cases. However, there is no specific clinical or radiologic signs. Therefore, a preoperative diagnosis is difficult. Bahadori and Leibow described its pathologic characteristics in 1973 and differentiated it from a variety of other lesions. Pulmonary inflammatory pseudotumors demonstrate a mixed inflammatory infiltrate with a preponderance of plasma cells, which vary in size and nuclear numbers. Variable numbers of small and transformed inflammatory cells as well as foamy cells are seen. A number of researchers believe that the inflammatory pseudotumor is in fact a regular inflammatory process which follows an interstitial pneumonia. Then, it transforms into an organized pneumonia and eventually, to an inflammatory pseudotumor. As many as 1/3 of the lesions start after a respiratory infection. The mechanism underlying transformation of inflammation to plasma cell granuloma has still not been elucidated clearly. In one pediatric case, production of cytokines, interleukine-6, and interleukine-beta has been demonstrated in blood and lung tissue. Another hypothesis, based on detailed histopathological studies, is that at least some plasma cell granuloma are slow-growing mesenchymal tumors with secondary inflammatory changes. It must be differentiated from such lesions as pseudolymphoma, malignant lymphoma, sclerosing hemangiomia (the last two have been erroneously considered to be the same entities as inflammatory pseudotumors), hamartoma, and chronic pneumonitis by histochemical staining. The inflammatory pseudotumors are not limited to the lung and can involve other systems.

Figure 3. Macroscopic appearance of inflammatory pseudotumor of the lung in a patient; A) a mass with a rubbery consistency and well-defined margin (diameter: 6.5 cm); B) cut section showing an encapsulated yellow-brown solid mass.

Figure 4. Clubbing of the fingers in one of the patients.
Figure 5. Microscopic appearance of inflammatory pseudotumor of the lung in a patient; A and B) mature plasma cells were the major component of the cellular population among bundles of fibroblasts or within collagen and hyaline membrane. Intermingled among the plasma cells are larger elements with characteristics of reticuloendothelial cells, inflammatory cells, and proliferation of collagen and foamed plasma cells (top X10) (bottom X40); C) alveolus in the peripheral mass with a tumor and inflammatory cells are seen (X40).

Multiple or bilateral nodules are rarely seen in the lung.

No organism has been ever found in histological and biochemical cultures. A wedge resection is usually sufficient in most cases of inflammatory pseudotumors of the lung. However, when a malignant lesion is suspected, a lobectomy or even pneumonectomy should be performed.

Locally invasive forms of the inflammatory pseudotumors have been described by many pathologists. Invasion of the surrounding tissues is present in these varieties and patients are often symptomatic. They may develop fever, dyspnea, fatigue, chest pain, and weight loss. Such cases often require more extensive excisions.

Recurrence after resections is rare, as has been the case in our patients.

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References

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