
Inflammatory Pseudotumor of the Lung Associated with Anemia: A Case Report and Review of the Literature

İrfan YALÇINKAYA*, İsmail DEMİRTAŞ**, Hayati AYAKTA***,
Nusret AKPOLAT****, İmdat DİLEK***, F. Hüsniye DİLEK****

- * Department of Thoracic Surgery, School of Medicine, Yüzüncü Yıl University Hospital, Van, TURKEY
** Department of Pediatric Surgery, School of Medicine, Yüzüncü Yıl University Hospital, Van, TURKEY
*** Department of Hematology, School of Medicine, Yüzüncü Yıl University Hospital, Van, TURKEY
**** Department of Pathology, School of Medicine, Yüzüncü Yıl University Hospital, Van, TURKEY

ABSTRACT

Inflammatory pseudotumor of the lung is a benign tumor which is rarely seen. A mass in the left lung was observed in the chest roentgenogram of an 8 year-old boy while investigating for anemia. Preoperative investigations could not render the exact diagnosis. Pneumonectomy was required in exploratory thoracotomy. Histopathologic diagnosis was fibrohistiocytic type of inflammatory pseudotumor. Four months after the operation, anemia was cured without any further treatment.

Key Words: Inflammatory pseudotumor, Lung, Anemia.

Turk J Haematol 2000;17(1):37-40.

Inflammatory pseudotumor is an often forgotten entity in the differential diagnosis of masses in the lung, despite the fact that, they are the most common benign primary pulmonary growths in children^[1-3]. Inflammatory pseudotumor is a broad histological term including organizing pneumonia type, fibrohistiocytic type and plasma cell granuloma type^[4] depending on the predominant cellular element. In 1939 Brunn^[5] reported an example of this lesion in a five year-old girl. In reports, up to 35 percent of them have occurred in

children less than 15 year-old^[2,6] as in our case. It is a rarely seen non-neoplastic mass and underlying pathogenesis is unclear^[6,7]. Although it is generally regarded as a reactive process, Matsubara et al. have suggested that it originates as an organizing intraalveolar pneumonia^[8]. In most chronic infectious and inflammatory diseases, anemia with hemoglobin ranging between 7 and 11 g/dL develop, where 20% of this anemia is characterized by hypochromic microcytic red blood cells^[9,10]. Only a few cases of inflammatory pse-

udotumor of the lung associated with anemia was reported in the literature^[6,7,11], but none of them had suggested whether the anemia improved postoperatively or not.

In this case, an association between inflammatory pseudotumor and anemia and a good outcome following surgical treatment is reported.

CASE REPORT

An eight-year-old boy, intermittently complaining of coughing, fever, fatigue and malaise, had previously taken treatment for anemia. There was no history of hemoptysis, weight loss or gain, easy bruising or bleeding. He was hospitalized to investigate a detected mass in his left lung, during the investigation for anemia. Physical examination of the chest revealed matity, and the respiratory sounds were decreased on auscultation. Computerized tomography of the chest showed a solid mass with sparse calcifications, involving whole lung fields of the left lung except anterior segment of upper lobe and lingula (Figure 1). Abdominal ultrasonography and bronchoscopic examination were normal. Laboratory tests revealed a high sedimentation rate of 70 mm/h, hemoglobin 8.1 g/dL, hematocrit of 26%, MCV of 58.9, MCH of 18.1, MCHC of 30.8 and RBC of 4.48. Others were in normal range. In the diagnostic and therapeutic thoracotomy, the left lung was completely solid except anterior and superior segment of lingula. There were also many hypertrophic lymph nodes

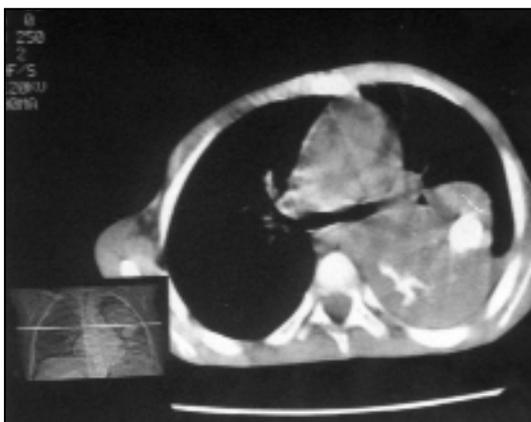


Figure 1. Tomographic appearance of inflammatory pseudotumor in the left hemithorax

des in the hilar and mediastinal areas. Left pneumonectomy and radical lymph node dissection was performed in spite of benign frozen section results, because the lesion covered almost all the lung. No complication was seen postoperatively. Hemoglobin and hematocrit values were found 14 g/dL and 43.3%, respectively without any further treatment four months after the operation.

Gross examination of the specimen revealed a 10x7x6 cm circumscribed mass in the left lung. The cut-surface of the lesion was quite homogeneous and grayish-yellowish-white in color. The consistency of the lesion varied from very hard to soft. On histopathological examination, the principal components of the lesion were spindle-shaped fusiform cells. They were arranged in a whorled, interlacing or storiform pattern. No cellular atypia or mitotic figures were found in these cells. Lymphocytes, histiocytes and plasma cells were present but not numerous. Interstitial eosinophilic amorphous hyaline material was found in some areas associated with focal calcification (Figure 2). Some epithelial-lined glandular structures, probably representing residual airspaces were seen. These histopathological findings were consistent with fibrohistiocytic type of inflammatory pseudotumor.

The patient is now quite healthy, 20 months after the operation.

DISCUSSION

Inflammatory pseudotumors are the most common benign primary pulmonary growths in children, however are uncommon benign tumors in the thoracic surgical literature^[1-3]. None of the laboratory data reviewed was helpful in making or suspecting the diagnosis of inflammatory pseudotumor, but elevation of the sedimentation rate, as in our case, and the aspartate aminotransferase level, as well as polyclonal gammopathy and thrombocytosis has been described^[7,12,13]. In a study of 91 cases, 10 cases showed hemoglobin less than 10 g, and high sedimentation rate was found in 13 of the patients^[6]. In another study of 7 cases, slight anemia was observed in three patients^[7]. Both values were in pathologic levels in our case. However, the results of the study of Berardi et al.

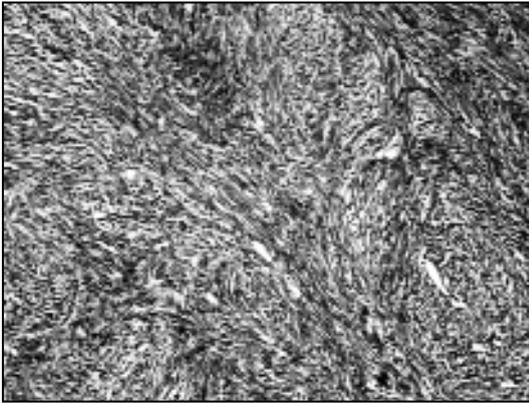


Figure 2. Microscopic appearance of the case, showing interlacing fibroblastic proliferation with a storiform pattern, admixed with chronic inflammatory cell infiltration. HE stains x 25

do not support the importance of these findings^[6].

Most of the patients have no symptoms^[6], but the clinical manifestations include fever, chest pain, recurrent pneumonia^[8,12,14], and about one third of the patients have history of an antecedent respiratory tract infection^[2,8]. Matsubara et al. reviewed the surgical pathology files of the Massachusetts General Hospital, Boston covering a 16 year interval (1970 to 1986) to find all lung nodules that were diagnosed as inflammatory pseudotumor^[8]. They found 32 such cases. Symptoms due to the tumor were present in 13 cases (40%), three patients complaint of chest pain, three patient gave a history of recurrent pneumonia and two patient had cough and two developed sudden hemoptysis. In our case there was cough, fever, fatigue and malaise which were all non-diagnostic signs and symptoms.

What makes our case more interesting is the improvement of anemia without any medical treatment on the 4th month after the operation. Pathogenesis is not clear in anemia due to chronic diseases; but shortening in life periods of red blood cells, disturbance in iron metabolism and increase in erythrocyte formation as insufficient compensation lead to anemia^[9-10]. Expected increase does not occur in erythropoietin levels in response to anemia. Cytokins such as tissue necrosis factor, gamma interferon, interleukin-6 and interleukin-1

beta are inflammatory, and seems to inhibit erythropoietin production^[15-17]. Spontaneous recovery of hypochrom microcytic anemia which is resistant to treatment, in our case after surgical excision, showed that the anemia was due to inflammatory pseudotumor.

It is generally agreed that the most appropriate approach of inflammatory pseudotumors of the lung should be surgical removal both for diagnosis^[7,8] and for treatment^[18,19]. Other resections including pneumonectomy can be applied according to the extension of the lesion. Radiation therapy has been tried for inoperable lesions with variable success^[20,21]. Although usually resected, inflammatory pseudotumors have been successfully treated with antibiotics^[22], corticosteroids^[23] and may regress spontaneously. Percutaneous needle aspiration biopsy is rarely helpful, since malignant tumors may be surrounded by inflammation. The prognosis of the patients with inflammatory pseudotumor is generally excellent, so was our case.

In conclusion, we suggest that hematologic abnormalities such as anemia could be seen in inflammatory pseudotumor of lung and cured without any further medical treatment after removal of the mass.

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Address for Correspondence:

İrfan YALÇINKAYA, MD

Department of Thoracic Surgery,
School of Medicine,
Yüzüncü Yıl University Hospital,
65200, Van, TURKEY