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Review of 30 pulmonary hamartoma cases: Follow-up and treatment

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The study protocol was approved by the Kocaeli Univercity Ethics Committee (11.9.2020 /2020-15.11).
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No conflict of interest was declared by the authors.

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Abstract

Background/Aim: The most common benign tumor of the lung is hamartoma. Lung hamartomas are tumors of benign mesenchymal origin with intraparenchymal or endobronchial localization. They are more common in the 40-60-years age range and among males. It is important to distinguish hamartomas from malignancies due to their radiological appearance. The main treatment is surgical excision. They tend to grow slowly, and malignant transformation is rare. Therefore, the decision of resection or follow-up and timing of the surgical intervention are important. In our study, we aimed to present our follow-up and treatment results and recommendations of 19 years of pulmonary hamartoma experiences.

Methods: In this case series, the data of 30 patients with hamartomas as diagnosed by histopathological examination, who underwent surgical resection, bronchoscopy, and transthoracic lung biopsy between January 2001 and May 2020 were analyzed retrospectively. Radiologic and nuclear medicine imaging features, which are clinically important, were divided into groups and compared.

Results: The mass was removed in 12 (40%) patients by enucleation, in 5 (16.7%) patients, by wedge resection, in 3 (10%) patients, by bronchoscopy, and in 1 (3.3%) patient, by bronchotomy, segmentectomy and lobectomy. Seven (23.3%) patients were diagnosed with tru-cut biopsy and did not undergo total mass excision. Trans-thoracic fine needle biopsy (TTFNB) was performed in 7 (23.3%) patients and histopathological diagnosis could not be obtained in any of these biopsies. Concomitant with hamartomas, lung squamous cell carcinoma, lung malignancy in the contralateral lung, prostate carcinoma, and leiomyoma were found in one patient (3.3%) each.

Conclusion: Because of their slow growth and rare malignant transformation, lung hamartomas should be diagnosed with less invasive methods. The patients should be followed carefully unless they are symptomatic.

Keywords: Hamartoma, Pulmonary hamartoma, Pulmonary nodule

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Introduction

The most common benign tumor of the lung is hamartoma [1, 2]. Lung hamartomas are tumors of benign mesenchymal origin with intraparenchymal or endobronchial localization [1]. They contain different components and are named according to the dominant tissue [2]. Although some genetic factors are suspected etiologically, no definite risk factors have been identified [3, 4]. The incidence of lung hamartoma is 0.025% –0.32% [1, 5]. They are more common in the 40-60-year age range and among males [2]. Their diameter is usually less than 2-3 cm [1], so they are often asymptomatic and detected incidentally [1, 2]. The main treatment is surgical excision [5]. They tend to grow slowly, and rarely undergo malign transformation. Therefore, the decision of resection or follow-up and timing of the surgical intervention are important.

In our study, we aimed to present our follow-up and treatment results and recommendations by sharing our 19-year experience with pulmonary hamartomas.

Materials and methods

Patient selection criteria and general features

The data of 31 patients with histopathological diagnoses of hamartoma who underwent surgical resection, bronchoscopy, transthoracic lung biopsy at Kocaeli University Hospital Department of Thoracic Surgery between January 2001 and May 2020 were reviewed retrospectively. The data used in our study were accessed through hospital files, hospital radiological imaging systems, and central records (e-Nabız, Mernis etc.). One patient with a diagnosis of hamartoma outside the lung parenchyma was excluded from the study. Finally, 30 hamartoma patients with sufficient data were included. Age, gender, smoking, illness histories, symptoms, tumor size-localization, radiological features, surgical technique, transthoracic biopsies, and accompanying malignancies were analyzed. Radiologic and nuclear medicine imaging features (calcification, fat density, diameter size, FDG uptake) were noted.

Approval 2020/262 was received from Kocaeli University Faculty of Medicine Ethics Committee on 11/09/2020.

Surgical features

All admitted patients underwent Thoracic Tomography (CT). Blood tests (hemogram, coagulometry, biochemistry), pulmonary function test (PFT), and echocardiography (EKO) were performed. The size of the tumor was determined by pathological examination in resected tumors and from the thorax CT images in non-resected ones. Those localized in one third of the medial hemithorax or associated with the mediastinal pleura were considered central tumors, while others were noted as peripheral.

Bronchoscopic or transthoracic biopsy were performed preoperatively in some patients to obtain a diagnosis. Patients diagnosed before the operation were not operated. Punch biopsy was performed in endobronchial lesions in bronchoscopic biopsy, while fine needle and tru-cut biopsies were performed in transthoracically.

Operations by posterolateral thoracotomy were performed to provide histopathological diagnosis and treatment. The operation type depended on the experience of the surgeon performing the operation. Radiological appearance features, lack of uptake on Positron emission tomography (PET-CT), and frozen pathological examination were effective in this decision. Enucleation was performed in lesions where the entire mass could be delivered, wedge resection was done in other patients, and lobectomy was performed in 1 patient due to concomitant primary lung malignancy. In enucleation, the tissue was repaired with direct sutures, while linear tissue staples were used in wedge resections. One drainage tube was inserted in all operations.

Postoperatively, patients were followed up by chest radiography and tube thoracostomy. Tubes that had no air leakage for more than 24 hours and whose daily drainages were less than 100 cc were withdrawn.

Follow-up

Routinely, a physical examination was performed, and chest x-rays were obtained at the 1st and 3rd weeks after discharge. Patients who were properly followed-up by us or whose health records could be found in the central recording system (e-Nabız) were included in the study.

Statistical analysis

All statistical analyses were performed using IBM SPSS for Windows version 20.0 (SPSS, Chicago, IL, USA). Kolmogorov-Smirnov and Shapiro-Wilk's tests were used to assess the assumption of normality. Continuous variables were presented depending on normal distribution with either mean (standard deviation) or (in case of no normal distribution) median (25^{th}-75^{th} percentile). Categorical variables were summarized as numbers (percentages).

Results

The mean age of the patients was 58.63 (12.08) years. Twelve (40%) patients were in the 40-60 age range and 18 (60%) were in the other age groups. Twenty-two (73.3%) patients were male and 8 (26.7%) were female. The median follow-up time was 5.76 (1.92-9.58) years. General demographic features are presented in Table 1 and hamartoma features are presented in Table 2.

Table 1: General characteristic

	Total (n=30)
Age (mean (SD)) (year)	58.63 (12.08)
Sex (n,%)	
Male	22 (73.3%)
Female	8 (26.7%)
Smoking History (n,%)	84(84.8%)
Disease History (n,%)	
Absent	16 (53.3%)
Pulmonary	1(3.3%)
Cardiovascular	5 (16.7%)
Other	8 (26.7%)
Symptom (n,%)	
Asymptomatic	20 (66.7%)
Cough	2 (6.7%)
Chest pain	8 (26.7%)

n: number, SD: Standard Deviation

Among the patients included in our study, transthoracic fine needle and tru-cut biopsies were performed in 7 (23.3%) patients each. Histopathological diagnosis was obtained in all patients who underwent tru-cut biopsy and none who underwent fine needle biopsies. Of these tumors diagnosed with tru-cut biopsies, 3 (42.9%) had calcification, 3 (42.9%) had fat density, and 4 (57.1%) had a diameter of 3 cm or more. The mean tumor size was 2.54 (0.85) cm. PET-CT was performed in 6 out of these 7 patients and 4 (66.67%) had no F-18 fluoro-2-deoxy-glucose

(FDG) uptake, 1 (16.67%) had no significant FDG uptake, while 1 (16.67%) showed significant FDG uptake.

Table 2: Tumor features

	Total n=30	
Localization n (%)		
Right upper lobe	4 (13.3%)	53.3%
Right middle lobe	3 (10%)	
Right lower lobe	9 (30%)	
Left upper lobe	8 (26.7%)	46.7%
Left lower lobe	6 (20%)	
Central / Peripheral n (%)		
Central	4 (13.3%)	
Peripheral	26 (86.7%)	
Tissue (n,%)		
Lung Parenchyma	26 (86.7%)	
Bronchial	4 (13.3%)	
Radiological Features n (%)		
Calcification	11 (36.7%)	
Fat Density	13 (43.3%)	
Tumor size n (%)		
<3 cm	6 (20%)	
≥3cm	24 (80%)	
Mean (SD) (cm)	1.98 (0.86)	
PET-CT n (%)		
Absent	15 (50%)	
No up-take	10 (66,7%)	
SUV-max < 2.5	2 (13.4%)	
SUV-max > 2.5	3 (20%)	

PET-CT: Positron Emission Tomography, SUV-max: Maximum standardized uptake value

The mass was removed in 12 (40%) patients by enucleation, in 5 (16.7%) patients, by wedge resection, in 3 (10%) patients, by bronchoscopy, and in 1 (3.3%) patient, by bronchotomy, segmentectomy and lobectomy. Total mass excision was not performed in seven patients diagnosed with tru-cut biopsies. Frozen pathological examination was performed in 14 (60.87%) cases during the operations.

Concomitant with hamartomas, lung squamous cell carcinoma, lung malignancy in the contralateral lung, prostate carcinoma, and leiomyoma were found in one patient (3.3%) each.

Discussion

Hamartomas constitute 3% of all lung tumors [6] and 6% of solitary pulmonary nodules [4, 5]. It is common in the male gender and between the 40-60-year age range [5, 6]. In our study, 73.3% of the patients were male, 40% were between 40-60 years of age.

Most hamartomas originate from the parenchyma and are localized peripherally [2,5]. In the study of Haberal et al. [2] including 24 patients, all lesions were of parenchymal origin, more frequently in the right lung, and 71% were peripherally located. In another study, 85% peripheral location was reported [6]. Endobronchial hamartomas are reported in 3-19.5% [5]. In our study, hamartomas were often localized in the lung parenchyma and peripherally. It was bronchial in 13.3%, found more frequently (53.3%) in the right lung and most frequently in the lower lobe of the right lung.

Hamartomas are often asymptomatic [2, 5-7]. Symptomatic ones are usually those that are large enough compress the endobronchial or surrounding bronchial-vascular structures. The most common symptoms are hemoptysis, cough, shortness of breath, and chest pain [1]. In our study, patients were often asymptomatic, and the most common symptom was chest pain.

Their growth rate is slow [8]. Most are small, and only a few giant hamartomas have been reported [1]. Lesion sizes are usually less than 2-3 cm [1, 2]. In the study of Haberal et al. [2], the size of 54.2% were 3 cm and less. The mean lesion size was 2.3 cm in the study of Çaylak et al. [6]. In our study, the mean lesion size was 1.98 cm and 80% was less than 3 cm in diameter.

Chest radiography and thoracic tomography (CT) can be used to aid diagnosis, but definitive diagnosis is obtained by histopathological examination [1]. Parenchymal hamartomas are usually well-circumscribed nodules with calcification on chest radiography. Endobronchial ones may yield indirect signs such as increased aeration or atelectasis in a hemithorax. Calcification (especially popcorn) in the mass, high fat content (up to 50%), slow growth, peripheral localization and lack of invasion are findings suggestive of hamartoma on thorax CT images [1, 2, 5]. About 10-30% contain calcification, and 50% contain fat, which is pathognomonic [4, 6]. In our study, we found 36.7% calcification and 43.3% fat content, close to the rates in the literature. In endobronchial hamartomas, no significant activity is expected in PET-CT except for some hamartomas with low lipid ratio [2]. Due to the interval in which our study was conducted, PET-CT was not studied in some patients, and in those who underwent PET-CT, 66.7% did not have any FDG uptake, while 20% of the patients had SUV-Max over 2.5.

It has been reported that needle aspiration biopsy is often insufficient for diagnosis [2, 5, 6]. In their study involving 20 patients, Çaylak et al. [6], 2 patients underwent TTFNB but could not be diagnosed histopathologically. In another study, transbronchial or transthoracic needle aspiration was performed in 12 patients and 3 patients (25%) could be diagnosed [4]. Contrary to expectations, some studies reported 85% sensitivity [9] or 86% diagnosis rate [10] in needle biopsy. Hamper et al. [10] stated that the diagnostic thoracotomy rate would decrease significantly with needle biopsy. Some studies state the necessity of surgery for definitive diagnosis [2]. While TTFNB was performed in 7 patients in our study and histopathological diagnosis was not obtained, diagnosis was obtained with tru-cut biopsy in 7 patients. Considering the distribution of tumor characteristics of patients diagnosed with tru-cut biopsy, no statistically significant difference was found in PET-CT in terms of FDG up-take, or in Thoracic CT in terms of calcification or fat content of the mass. The number of patients with tumors larger than 3 cm was significantly higher among those who underwent tru-cut biopsy.

Bronchial carcinoma development is more common (7%) in patients with pulmonary hamartoma [6]. In two hamartoma studies with 24 and 39 patients conducted in our country, bronchogenic carcinoma association was reported in 2 patients each [11]. In one study, the risk of pulmonary malignancy in patients with hamartoma was 6.3 times higher than the general population [12]. In our study, there was 1 (3.3%) lung squamous cell carcinoma, 1 (3.3%) lung malignancy in the contralateral lung (6.6% in total), 1 (3.3%) prostate carcinoma, and 1 (3.3%) leiomyoma in patients simultaneously with hamartomas.

Surgical excision is sufficient in treatment [1]. Care should be taken to lose as little parenchyma as possible. Generally, enucleation or wedge resection is sufficient [5, 6]. Sometimes, large parenchymal resection or even pneumonectomy can be performed due to the hamartoma's location and its relationship with the surrounding tissue and symptoms [1]. Lobectomy rates between 6.7-26.3% and pneumonectomy rates between 1.5-13.3% have been reported [4]. Post-surgical mortality and morbidity is very low [2]. Forty percent of our patients underwent enucleation,

16.7% underwent wedge resection, 3.3% underwent lobectomy, and no pneumonectomy was performed. The indication for lobectomy was malignancy accompanying the hamartoma. No mortality or serious morbidity was observed in any of our patients.

Malignant transformation or recurrence has been reported very rarely [5]. In two studies which 15 and 20 patients underwent surgical treatment, recurrence [4, 6] or malignant transformation were not reported [6]. In a study that reported surgical resection in 39 patients, recurrence was reported in 1 (2.6%) patient, but no malignant transformation [5]. In another study on 52 patients, malignancy developed in the hamartoma excision site in 4 (7.69%) patients [12]. No recurrence or malignant transformation was observed in our follow-ups.

Since malignancy or recurrence may develop, who to follow and until when is another matter of discussion [5]. Some authors recommend surgery in young and middle-aged cases with a growing lesion or persistent symptoms [4, 5]. Guo et al. [5] reported that because hamartomas tend to grow and may cause malignancy by inducing chronic inflammatory processes, lesions larger than 2.5 cm, and solitary lesions which cannot be distinguished from malignancy, should be excised. In a study, resection was recommended for large lesions, those increasing rapidly in size, and symptomatic patients [7]. In another study, resection was recommended in young or middle-aged and symptomatic patients [9]. Some authors recommend follow-up in asymptomatic lesions proven by biopsy [7]. In our study, surgery was not performed in any of our patients due to their symptoms. In general, the procedure was performed for malignancy exclusion regardless of the size of the lesion (except size increase). We followed up 10 (33.33%) patients by diagnosing them with tru-cut biopsy or bronchoscopy.

The limitations of our study include low number of cases, although it was sufficient considering its incidence, and its retrospective nature. The study will expand with more case series in the future.

Conclusion

It is important to distinguish hamartomas from malignancy due to their radiological appearance. Because of their slow growth and low risk of malignancy, hamartomas may be followed closely until they become symptomatic. Their definitive treatment is surgical resection.

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