Rev Port Pneumol. 2017;xxx(xx):xxx–xxx

revista portuguesa de
PNEUMOLOGIA
portuguese journal of pulmonology
www.revportpneumol.org

ORIGINAL ARTICLE

The frequency of lung cancer in patients with pulmonary hamartomas: An evaluation of clinical, radiological, and pathological features and follow-up data of 96 patients with pulmonary hamartomas

G.H. Ekinci[a,*], O. Hacıömeroğlu[a], A. Ersev[b], L. Alpay[c], H. Özgen[a], A. Yılmaz[a]

[a] *Department of Pulmonology, Süreyyapaşa Chest Diseases and Thoracic Surgery Training and Research Hospital, Maltepe, İstanbul, Turkey*
[b] *Department of Pathology, Süreyyapaşa Chest Diseases and Thoracic Surgery Training and Research Hospital, Maltepe, İstanbul, Turkey*
[c] *Department of Thoracic Surgery, Süreyyapaşa Chest Diseases and Thoracic Surgery Training and Research Hospital, Maltepe, İstanbul, Turkey*

Received 19 October 2016; accepted 25 April 2017

KEYWORDS
Hamartoma;
Lung neoplasms;
Diagnosis

Abstract
Purpose: To investigate the frequency of lung cancer in patients with pulmonary hamartomas and to evaluate clinical, radiological, and pathological characteristics of pulmonary hamartomas.
Basic procedures: We reviewed pathology records of pulmonary hamartomas diagnosed between 2003 and 2014. Medical records and the hospital electronic database were also reviewed for each patient to obtain clinical, radiological, and pathological characteristics of pulmonary hamartomas and accompanying malignancies.
Main findings: Ninety-six patients with pulmonary hamartomas were identified. There were 26 females (27%) and 70 males (73%), with a mean age of 56.2 years (range 22–87 years). Malignancies were detected in 23 patients (24%), which developed previously in five patients (1 synchronous, 4 metachronous lesions), and concomitantly in 18 patients (with origin from the lung in 17 patients and from the pleura in 1 patient).

* Corresponding author.
E-mail address: gulbanuh@hotmail.com (G.H. Ekinci).

http://dx.doi.org/10.1016/j.rppnen.2017.04.008

Please cite this article in press as: Ekinci GH, et al. Rev Port Pneumol. 2017. http://dx.doi.org/10.1016/j.rppnen.2017.04.008

Principal conclusions Our results show that patients with pulmonary hamartomas may have coexisting lung malignancies.

Introduction

The term hamartoma was first introduced by Albrecht in 1904 to describe lesions that contain an abnormal mixture of tissue elements or an abnormal proportion of a single element, normally present in an organ. While hamartoma was initially considered a development malformation, it is now classified as a true benign mesenchymal tumor, consisting of cartilage, fat, fibromyxoid connective tissue, smooth muscle, and bone.[1,2] Hamartomas can occur in any organ or region. Pulmonary hamartoma is the most common benign tumor of the lung, accounting for 3% of all lung tumors.[3] Its incidence was found as 0.025% and 0.32% in two large autopsy-based studies.[4]

There have been many reports suggesting an association between hamartoma and malignancy,[5–10] and the likelihood of its malignant transformation to carcinoma or sarcoma.[5–7] The risk for lung cancer in pulmonary hamartoma patients was estimated to be 6.3 times as high as that expected for the general population after adjustment for age, sex, and ethnicity.[8] Additionally, patients with pulmonary hamartomas can develop synchronous or metachronous lung cancers.[9,10] The aim of this study was to investigate the frequency of lung cancer in patients with pulmonary hamartomas and to evaluate clinical, radiological, and pathological characteristics of pulmonary hamartomas accompanied by lung cancer.

Materials and methods

This retrospective study was conducted at Süreyyapaşa Chest Diseases and Thoracic Surgery Training and Research Hospital, İstanbul, Turkey, after being approved by the scientific committee of our institute. A comprehensive review of pathology records was made for pulmonary hamartomas diagnosed between 2003 and 2014, which yielded a total of 97 patients. All histologic slides were re-examined by a pathologist experienced in thoracic pathology to re-confirm the diagnosis of pulmonary hamartomas. One patient whose diagnosis could not be confirmed was excluded from the study. Thus, the study included 96 patients with pulmonary hamartomas.

Medical records and the hospital electronic database were reviewed for each patient and a standardized data-collection form was used to collect the following information: gender, age at diagnosis, history of smoking, medical history, the presence of malignancies (previous, concurrent), symptoms, radiological and imaging findings, location and size of hamartomas, findings of preoperative diagnostic investigations, duration between diagnosis of malignancy and pulmonary hamartoma, malignancy staging, treatment modalities, histologic features, duration of follow-up, and outcome. Follow-up information included data till January 2015.

Results

Of 96 patients with pulmonary hamartomas, 26 (27%) were females and 70 (73%) were males, with a female-to-male ratio of 1:2.7. The mean age was 56.2 years (range 22–87 years). Only one patient was younger than 30 years. Sixty-one patients (63.5%) were between 30 and 60 years and 34 patients (35.4%) were older than 60 years. With respect to smoking, 62 patients (64.6%) were smokers (mean 41.2 pack-years), and 34 (35.4%) had never smoked.

At presentation, 27 patients (28%) were symptom-free, while the remaining 69 patients (72%) had pulmonary and/or constitutional symptoms. The presenting symptoms were cough ($n = 27$), chest pain ($n = 26$), dyspnea ($n = 23$), hemoptysis ($n = 10$), fever ($n = 7$), sputum production ($n = 5$), and constitutional symptoms ($n = 7$). The duration of symptoms ranged from 4 days to 1 year.

Chest X-ray examination was performed in all patients, which showed abnormal findings in all but three patients. Eighteen patients also had findings associated with concomitant malignancies. All patients underwent computed tomography and bronchoscopic examinations. The diagnoses of pulmonary hamartomas were established via bronchoscopic biopsies in 19 patients, computed tomography-guided transthoracic fine-needle aspiration in three patients, and surgical biopsies in 74 patients. A single hamartoma was found in 93 patients (97%) which was located in the right lung in 54 patients (58%) and in the left lung in 39 patients (42%). Three patients (3%) had multiple hamartomas, with two patients having two hamartomas in the right and left lungs, and one patient having three hamartomas in the same lobe. The hamartomas were parenchymal in 72 patients (75%) and endobronchial in 24 patients (25%). The mean diameter of pulmonary hamartomas was 22.2 mm (range 3 mm–100 mm).

There was no evidence for malignancy in 73 patients (76%) including 50 males and 23 females, with a mean age of 53.9 years (range 22–76 years). Data on these patients are summarized in Table 1. Of these, 44 patients (60.3%) were smokers and 50 patients (68.5%) were symptomatic at presentation. The most common radiological pattern was a solitary pulmonary nodule, followed by a solitary mass. Seventy patients (95.9%) had a single hamartoma and three patients (4.1%) had multiple hamartomas. The hamartomas were parenchymal in 52 patients (71.2%) and endobronchial in 21 patients (28.8%). The diameter of pulmonary hamartomas ranged from 6 mm to 100 mm (mean 25.4 mm). ^{18}Fluoro-deoxyglucose positron emission

Please cite this article in press as: Ekinci GH, et al. Rev Port Pneumol. 2017. http://dx.doi.org/10.1016/j.rppnen.2017.04.008