Hamartoma of the Lung
A Clinical Study of 25 Cases

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Introduction

Hamartoma is an abnormality of growth first accurately described by Albrecht in 1904.1 This report will consider pulmonary lesions only, although this pathologic process may occur in other organs, notably the liver. Excluded from this report will be consideration of the large pulmonary lesions which occur in newborns and lead to rapid death of the patient.2 We had no such case in our series and indeed there is some speculation as to whether this type of tumor properly fits the category of pulmonary hamartoma.

Hamartomas of the lung occur principally in the peripheral pulmonary parenchyma although a few are found in the bronchi. They are more common in men and have no predilection for one lobe or another. Although pulmonary hamartomas are found principally in the fourth decade with some spread into the third and fifth decades, in our series the age distribution was from 16 to 76 years. Most commonly, hamartomas present as asymptomatic pulmonary lesions noted on radiologic examination of the chest.

The importance of hamartomas rests on their recognition in the differential diagnosis of pulmonary tumors and on the now well-documented fact that these tumors may grow while under observation and hence give concern to the clinician regarding malignancy. The absence of a clear cut clinical picture or radiologic image makes the definitive diagnosis of hamartoma by non-surgical means most difficult.

Case Material

This report deals with an analysis of a series of 25 clinical cases taken from the files of the Department of Thoracic Surgery and the Department of Pathology of the University Hospital from 1925 to 1961. During this period an additional 15 cases were discovered as incidental findings at necropsy; these, however, have been excluded from this study since we wished to consider only those cases which were of significance to the patient during life and which have been verified histologically. Six other cases encountered in our search through the records were not included because the diagnosis was on a clinical impression alone and had not been established by histologic examination.

From 1925 to 1961, there have been approximately 678,300 admissions to the University Hospital so that one surgically proved hamartoma has appeared for every 27,100 admissions thus indicating the rarity of these lesions. Of 7,972 necropsies at the Mayo Clinic 20 cases were found for an incidence of 0.25 per cent.3 In the English medical literature to date, a total of approximately 200 cases have been reported.

Because of the increased number of chest x-ray examinations being performed, more of these lesions. Of 7,972 necropsies at the section to the lesions being discovered, more of them are being resected because of the many improvements in the field of surgery. For the past three to four years, in our department, we have been seeing two to three cases a year.

Etiology

The etiology of hamartomas is unknown. Several theories have been proposed and considered by writers on this subject.4

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*From the University Hospital, University of Michigan, Ann Arbor.
The third theory of neoplasia of cartilaginous elements may be the pathologic mechanism. A histologic definition intervenes, since the description and appearance of the tumors we have encountered favor hyperplasia over new growth. Nevertheless, one aspect of this theory deserves attention since the spectre of malignancy lurks in the background of hamartomas. In two of our cases, proved malignancy occurred: in one case several years after a benign hamartoma had been removed from the same lung, and in another case a simultaneous adenocarcinoma and hamartoma existed in adjacent areas of a resected lung specimen. We have encountered one other case which was not included in our series, but which was originally considered by the pathologist to be a fibrosarcoma of the lung arising from a hamartoma. After consideration of all microscopic aspects, this case was not included, but sufficient evidence was present to raise a question as to the role of the neoplasia.

The fourth theory is that these tumors arise in response to inflammation. We have not been able to demonstrate any previous pulmonary parenchymal irritation except in two patients, one of whom had silicosis and who, while under observation, developed a discrete nodule which, when resected, was a hamartoma. The second patient had a pulmonary resection for tuberculosis, and in addition to the tuberculosis a nodule was found which was a hamartoma. It would seem that if this theory were operative more clinical evidence of infection would be found. Also, since pulmonary infection is common we should see more hamartomas.

**Clinical Features**

Hamartomas of the lung have occurred more commonly in men than in women in most reported cases. In our own series, we have found 17 men and eight women with this tumor.

While our age range was wide, from 14 to 76 years, the distribution was in the fourth decade with spread into the third...
and fifth decades. We did not find any cases in infants with a large irregular tumor presenting.

The majority of our patients' tumors were located in the pulmonary parenchyma as was true in most series. We had 23 cases with peripheral lesions, and two with endobronchial involvement; this corresponds to what several other authors have found. The lesions were uniformly spread throughout the pulmonary parenchyma with no predilection for any one lobe.

Six of our patients were symptomatic and 19 cases were asymptomatic. The symptoms of cough usually moderate to mild and non-productive, chest pain, fever, pneumonia, and hemoptysis all were recorded as individual or collective findings. Three patients complaining of chest pain had a contralateral lesion discovered on roentgenograms. The 19 asymptomatic cases were discovered by incidental x-ray examination of the chest. This was well exemplified by a chiropractor who served as a willing subject for the testing of a new piece of x-ray equipment for a colleague and was found to have a coin lesion which, on surgical removal, proved to be a typical hamartoma.

In reviewing the records of these patients, we have not encountered any common geographic, economic, or sociologic features which would serve as a clue or indication as to the origin of these tumors.

As with many pulmonary lesions, particularly of the size we are dealing with, physical examination was uniformly of no assistance in the evaluation of these patients.

Radiographic Findings

Hamartomas are seen on photofluorograms and standard roentgenograms of the chest. Special techniques are usually not required. They usually appear as well-circumscribed lesions in any portion of the lung field, circular in outline, and well demarcated from surrounding pulmonary parenchyma. Some radiologists have been able to detect a bosselated appearance on x-ray films although we have not been able to so in our series.

The usual size of the lesions is about 3.0 cm. in diameter; however, a 20 cm. lesion has been recorded by Hodges. In our series, the smallest lesion was 1.0 cm. and the largest 8.0 cm. in diameter, the majority being 1.5 to 3.0 cm.

Although calcification of these lesions has been encountered somewhat more frequently in other series, only one of our cases demonstrated it.

Laminography has been used in some of our more recent cases and, other than delineating the lesion better, has not been of assistance in establishing the diagnosis. It has generally been of value in demonstrating the presence of calcium in lesions and because of the absence of calcium in hamartomas, has served as further indication for surgical intervention.

In the two cases of endobronchial hamartomas, the lesion itself was not visualized, but there was radiographic evidence of peripheral atelectasis which, coupled with hemoptysis, led to bronchoscopy and the identification of the disease process.
surprising when one considers the peripheral location of the tumors and their lack of bronchial communication.

**Therapy**

All of our cases were proved at surgery, since this was one of the defining features of our study. This is the only method of making proper histologic diagnosis during life, although it can be suspected on other grounds.

Local excision including such procedures as enucleation, and wedge excision was accomplished in 17 cases. One standard segmental resection and six lobectomies were carried out. One pneumonectomy was performed in a patient with pulmonary tuberculosis for which the resection was done, and the hamartoma was an incidental finding on sectioning the specimen.

The tumor in one patient was initially treated by bronchoscopic resection which subsequently was found to be inadequate, and the patient had a right middle and lower lobectomy four months later.

Another case was of interest surgically in that bronchoscopy was not adequate for removal of the tumor. Later at thoracotomy, a bronchotomy was utilized to resect the tumor. This proved to be inadequate because left upper lobe atelectasis

**Table 1**

<table>
<thead>
<tr>
<th>Description</th>
<th>Frequency</th>
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<tbody>
<tr>
<td>Number of Cases</td>
<td>25</td>
</tr>
<tr>
<td>Men</td>
<td>17</td>
</tr>
<tr>
<td>Women</td>
<td>8</td>
</tr>
<tr>
<td>Site of Lesion</td>
<td></td>
</tr>
<tr>
<td>Peripheral</td>
<td>23</td>
</tr>
<tr>
<td>Endobronchial</td>
<td>2</td>
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<tr>
<td>Age Range</td>
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</tr>
<tr>
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</tr>
<tr>
<td>Median age</td>
<td>4th decade</td>
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<tr>
<td>Symptoms</td>
<td></td>
</tr>
<tr>
<td>Asymptomatic</td>
<td>19</td>
</tr>
<tr>
<td>Therapy</td>
<td></td>
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<tr>
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<td>Lobectomy</td>
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</tr>
<tr>
<td>Pneumonectomy</td>
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</tbody>
</table>

Bronchography has not been used commonly in the investigation of these tumors because of their peripheral location. Experience has shown that bronchograms are not useful in evaluating coin lesions as most of these are. Only one case in our series had bronchography and this showed failure of filling of the superior dorsal segment of the left lower lobe where the tumor was located.

A most important point when considering the radiographic findings in hamartomas is the fact that these lesions can enlarge in size when followed with serial roentgenograms. A lesion in one of our patients enlarged over a period of nine months, another enlarged over a period of six years, and a third enlarged over a period of four years. Weisel et al. reported ten cases of hamartoma, all of which were known to have enlarged radiographically during varying periods of observation. The knowledge that these lesions may enlarge is important and is another indication for surgical intervention.

**Bronchoscopy**

Bronchoscopy has been carried out in 16 of our 25 cases. In the two cases of endobronchial hamartoma this revealed the true nature of the lesion; in the remaining 14 cases it was not contributory. We have examined by cytologic methods the sputum, which was obtained at bronchoscopy, and have noted no cellular abnormality to aid in the diagnosis. This is not

**Figure 3:** Cut surface of a hamartoma which was bosselated, yellow-white and shelled out of the surrounding lung substance.
postoperative course.

The type of resection at surgery is dictated by the location of the lesion, the size of the lesion, and the surgeon's awareness of the possibility of the tumor being a benign lesion. If the hard, circumscribed nature of the tumor indicates to the surgeon the possibility of hamartoma, he will endeavor to use local excision. If, however, roentgenographic appearance and growth make the surgeon consider malignancy more strongly, a lobectomy should be carried out.

**Gross Appearance**

Hamartomas are spherical to ovoid in contour and have a smooth or bosselated surface. They generally delineate well from surrounding pulmonary tissue except for endobronchial lesions. One of our endobronchial lesions was pedunculated and easily extracted, while another had produced distal atelectasis and necessitated further resection as noted previously.

In our series, the lesions ranged around 3.0 cm. in diameter although as noted previously larger sizes have been recorded. On palpation, the tumors are firm and rubbery. They will shell out of surrounding parenchyma. The sectioned surface is yellow-white in color, the exact shade being a result of the combination of cartilaginous and lipomatous tissue present. The tumors cut with the consistency of cartilage and occasionally, although uncommonly in our series, calcified flecks may be seen.

**Microscopic Appearance**

Hamartomas consist primarily of hyaline, fibrous, or elastic type cartilage. Dense hyaline connective tissue may be present with fat almost invariably present. Clefts and cystic spaces lined by a respiratory type of epithelium are also a feature of this tumor.

The tumors do not have any evidence of malignancy although as mentioned two of our cases had an associated malignancy.

**Follow-up**

The 25 cases have been followed for varying periods ranging from several months to eight years. Other than the appearance of a squamous cell carcinoma five years after the hamartoma removal in one patient, the others have done well with no further evidence of pulmonary disease.

**Discussion**

This series of 25 clinical cases has been evaluated since each of them presented to the surgeon a diagnostic problem which was ultimately resolved by surgery and histologic proof of the diagnosis. We have
not included our series of 15 necropsy cases with this group, since these tumors were incidental findings at the time of necropsy and did not cause any concern to the surgeon or the patient during life. In addition, we have found in our records six additional cases which presented sufficiently strong evidence for a clinical diagnosis of hamartoma, but in which no surgery was performed. Four of these were in older patients in whom the risk of resection was too great. Two were discovered to have had a stable lesion for many years and observation was continued. While most of these probably represent hamartomas, we did not include them because of lack of histologic verification.

The clinical features of hamartomas have been apparent in this group. They are more common in men and in the older age group. They are generally peripheral in location and may appear anywhere in the lung fields. The two cases of endobronchial hamartomas that we had in our series were of interest in that both of them presented with hemoptysis and yet this symptom was absent in the 23 other cases.

The asymptomatic nature of these tumors is not surprising because they are usually peripheral, well delineated from lung parenchyma, and in the reported cases rarely have produced compression of vital structures to give any symptoms.

Their x-ray appearance is that of a coin lesion, frequently, and hence they should be considered in a differential diagnosis of coin lesions.*

Of great importance is the fact that these tumors may increase in size under observation. This has been demonstrated in our series as well as others.

The infrequency of calcification in these lesions is a stimulus for surgical extirpation.

The surgical approach to these tumors is thoracotomy followed by a procedure dictated by the surgeon’s evaluation of the age of the patient, the x-ray appearance of the lesion, the gross characteristics of the tumor, and the extent of involvement of the lung. It has been evident to us that the surgeon in many instances has made the correct diagnosis on inspection and has limited the removal to a wedge excision or enucleation. In other instances there was a good deal of indecision culminating in a more major resection. The pathologist, however, by frozen-section methods can usually quite easily establish the diagnosis of hamartoma.

We have not been able to detect a single sign, symptom, finding, or even a combination of these to make an unequivocal diagnosis of hamartoma short of resection and microscopic diagnosis. The occupational background of our patients has varied widely from a history professor to a pickle manufacturer, chiropractor, machinist, and a housewife. It is most important to recognize this lesion in the differential diagnosis of coin lesions.

**Summary**

A series of 25 clinical cases of pulmonary hamartomas and their significant features has been presented. These usually appear in an asymptomatic patient with a lesion noted on x-ray anywhere in the lung parenchyma, appearing in men in the 40 to 50 year age group. The gross appearance of a firm yellow-white lesion which microscopically is composed of cartilage with an admixture of fat and epithelial clefts is demonstrated. The surgical therapy of the tumors usually consists of local excision. The long-term results in this type of tumor are excellent.

**Resumen**

Se ha presentado una serie de 25 hamartomas pulmonares, así como sus características importantes. Generalmente aparecen en una persona sin síntomas, en la que se descubre la neoplasia a los rayos X en cualquier parte del pulmón, en hombres de 40 a 50 años de edad. La apariencia macroscópica es de una formación blanco-amarillenta que se compone microscópicamente de cartílago con mezcla de grasas y hendeduras epiteliales. El tratamiento quirúrgico es la excisión local. Los resultados a largo plazo son excelentes.
Les auteurs présentent un groupe de 25 cas cliniques d'hamartomes pulmonaires avec leurs traits principaux. Ils apparaissent généralement chez un malade ne présentant aucun symptôme atteint d'une lésion découverte radiologiquement, dans n'importe quelle zone du parenchyme pulmonaire. Il s'agit d'hommes âgés de 40 à 50 ans. L'apparence macroscopique est celle d'une lésion dense jaune-bleanche, qui est microscopiquement composée de cartilage avec pénétration de graisse et de fentes épithéliales. Le traitement chirurgical des tumeurs consiste habituellement en leur exérèse locale. Les résultats à longs termes de ce type de tumeur sont excellents.

ZUSAMMENFASSUNG

REFERENCES

For reprints, please write Dr. McElvein at 2134 Nicholausville Road, Lexington, Kentucky.

BRONCHOESOPHAGOSCOPY POSTGRADUATE COURSE
The Department of Laryngology and Bronchoesophagology, Temple University, will present two courses in bronchoesophagology: October 7-18, 1963, and March 9-20, 1964. Tuition for each course is $250. Application and further information may be obtained by writing to: Chevalier Jackson Clinic, Temple University Medical Center, 3401 North Broad Street, Philadelphia 40, Pennsylvania. The course will be given by Drs. Charles M. Norris, Gabriel F. Tucker, Jr. and Walter H. Maloney.

RESULTS OF MAJOR SURGERY AND SHORT-TERM CHEMOTHERAPY
An outline of the surgical work for pulmonary tuberculosis in the Cheshire Joint Sanatorium over a five-year period from 1952 to 1957 has been described. There was a three to eight year follow-up of 96.6 per cent of the cases. There were 418 patients. The mortality was 0.48 per cent in the sanatorium. The relapse rate was 2.1 per cent. No patient has resistent organisms. It is concluded that surgery was a powerful adjunct to short-term chemotherapy in the treatment of pulmonary tuberculosis. Such a combination should be useful where long-term chemotherapy is impracticable, particularly in underdeveloped countries.


LOCALIZED AIR TRAPPING PULMONARY DISEASE
The benefit of pulmonary resection for various forms of air trapping pulmonary disease has been confirmed by preoperative and postoperative studies of pulmonary function in 24 patients. The removal of presurized air trapping pulmonary processes results in a decrease of the total lung capacity, decrease of the residual volume relationships to the total lung capacity, decrease in the nitrogen retention, and a remarkable increase in the maximum breathing capacity in a significant number of cases. The removal of the presurized processes also results in a decrease in the parenchymal compression, a correction of the mediastinal shift, a decrease in diaphragmatic immobilization, and allows the diaphragm to cup, or elevate its center, and facilitate the ventilatory dynamics of the lung and chest wall. The decrease in the nitrogen retention indicates that the increase in ventilatory dynamics also facilitates the mixing and exchange of gases of the residual air and, in some cases, has resulted in the correction of longstanding respiratory acidosis.