Hamartoma Simulating Ipsilateral Metastasis in a Case of Primary Bronchogenic Carcinoma

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Although primary carcinoma of the lung is fairly common, representing about 10 per cent of all carcinoma cases seen at autopsy, the co-existence of a benign pulmonary hamartoma must be rare. Fortunately, in the case to be presented, the two lesions were in the same lung so that pneumonectomy removed both tumors. Had the benign lesion been in the contra-lateral lung, it would have been considered to be a metastasis from the bronchogenic carcinoma and the patient would have been denied the benefit of surgery.

Case Report

D.K. Case 144698. A 71 year old white male was admitted to the Thoracic Surgical Service of the Hospital for Joint Disease on March 27, 1951 with a history of having had productive cough and fever three weeks prior to admission. At that time, physical examination revealed signs of pneumonia of the lower lobe of the left lung which was treated with penicillin. The symptoms disappeared but roentgen examination revealed a residual infiltration lesion of the left upper lobe which was suggestive of pulmonary neoplasm and hospital admission was recommended.

Past History was negative except for benign prostatic hypertrophy which was treated at another hospital two years previously by supra-pubic prostatectomy. Upon admission, he presented no weight loss, hemoptysis or expectoration.

Physical Examination revealed dullness to percussion of the left chest down to the level of the third thoracic spine with diminished breath sounds and vocal and tactile fremitus. No adenopathy was present. The remainder of the examination was negative.

Laboratory examinations of urine, blood, non-protein nitrogen, total protein, inorganic phosphatase, alkaline phosphatase, acid phosphatase and chlorides and hemogram were all within normal limits.

Roentgenograms of the chest taken April 4 (Figure 1) and laminograms on April 7, 1951 (Figure 2) showed a large globular area of infiltration extending from the hilum of the left lung into the apex at the level of the second intercostal space. In addition, a large fibrotic nodule or area of infiltration was noted in the plane of the fourth intercostal space. This was interpreted as probable bronchogenic carcinoma of the left upper lobe with adjacent pneumonitis and ipsilateral metastasis. Roentgenographic examination of the long bones was negative for metastases.

Bronchoscopy was performed on April 16, 1951 and revealed the trachea deflected to the right with visualization of the left upper lobe orifice not possible due to the anatomical arrangement. No infiltration, obstruction or neoplasm was noted within the range of bronchoscopic vision and no biopsy was taken. Papanicau smears from the left main bronchus showed no abnormal or atypical cells.

Sputum Examination by the Papanicau method, however, showed occasional clusters of deeply staining cells with hyperchromatic nuclei. These were considered as atypical cells but without malignant features.

Operation was performed on April 28, 1951 under endotracheal ether, cyclopropane anaesthesia in the "face-down" position. The sixth rib as well as a short posterior segment of the fifth were removed. The pleural space could not be
entered due to synechias between visceral and parietal pleurae so that an extra-pleural approach was used and a pleuro-pneumonectomy performed. After the apex was brought down by blunt and sharp dissection, the mediastinum was found to be flaccid and uninvolved except for some large, soft, edematous lymph nodes which were removed with the lung. Individual cotton ligatures were used on vessels, and five silk plication sutures on the bronchus in the manner of Overholt.\textsuperscript{2} The pleural cavity was thoroughly irrigated with saline and one tube dependent thoracostomy performed.Silk was used in layers for closure.

**Post-operative Course:** The drainage tube was removed after 24 hours. On the second post-operative day, he became slightly dyspneic and his skin was found to be cold and moist despite normal temperature and blood pressure. The pulse was grossly irregular and an electrocardiogram revealed many supraventricular premature contractions. He was digitalized with digoxin but maintained a tachypnea and fine moist rales at the right base for two weeks post-operatively (Figure 3). On May 30, 1951 he was discharged to a convalescent home where he remained for several weeks prior to returning home. He was seen at the Thoracic Surgical Clinic on October 18, 1951 where roentgenogram revealed obliteration of the left hemi-thorax by diffuse opacity without fluid level and only slight ipsilateral shift of the mediastinum; the right lung appeared normal. He had no complaints. There was no dyspnea on ordinary activity and the heart showed regular sinus rhythm. No lymph nodes were palpable.

**Specimen:** The lung was cut immediately after pneumonectomy, and unfortunately in doing so it was rendered unsuitable for photography. An irregular area of consolidation was present involving about two-thirds of the left upper lobe and extending into the apex. The upper lobe bronchus was patent for about 0.5 cm. and was occluded at this point by a friable whitish mass which infiltrated the bronchial wall. Upon pressure over the substance of the induration, a moderate amount of green, purulent material could be expressed. The indurated area extended almost to the visceral pleura at the periphery and contained numerous dilated bronchi and small saccular and cylindrical cavities which represented bronchiectasis and early abscess formation. The surrounding pulmonary parenchyma was atelectatic, firm and showed marked evidence of inflammatory exudate, which extended to the interlobar fissure and the pleura. In the area of the lingula and 0.5-0.7 cm. below the anterior visceral pleura was a round 1.3 by 1 by 1.2 cm. circumscribed nodule of whitish semi-gelatinous tissue which could be enucleated from the lung parenchyma with ease. The surrounding pulmonary parenchyma was not adherent and was smooth after the nodule was enucleated. The nodule itself presented a rather friable center surrounded by whorls of thickened whitish tissue. The central area was more gelatinous or mucoid than the peripheral portion.

**Microscopic examination** revealed a rather well differentiated epidermal carcinoma of the left upper lobe bronchus with atelectasis and acute and chronic inflammation of the peripheral lung parenchyma (Figure 4). The nodule was found to be composed of irregular strands and islands of cartilage interspersed with fat cells, fibroblasts, smooth muscle fibers and bronchial mucosa (Figure 5). This was considered to be a benign hamartoma.

**Discussion**

Warren and Gates\textsuperscript{3} found 6.8 per cent of 2,829 autopsies on patients with carcinoma disclosed multiple primary lesions. Cahan, Butler and Watson\textsuperscript{4} reported 20 cases of multiple primary carcinomata, one being a triple primary—breast, sigmoid and lung. Hochberg, Grayzed, Berson and Rosenberg\textsuperscript{1} reported a fascinating case in which a fibrosarcoma and carcinoma were present in one lung with a hamartoma in the other. In addition, there
**FIGURE 1**

*Figure 1:* Pre-operative roentgenogram showing infiltrating lesion left upper lobe and solitary nodular density (Arrows).

**FIGURE 2**

*Figure 2:* Pre-operative laminogram.—*Figure 3:* Roentgenogram on 12th post-operative day. Subsequent films show disappearance of fluid levels and subcutaneous emphysema.
were several benign tumors in six other organs. Unfortunately, no roentgenograms were published.

Albrecht in 1904 introduced the word “hamartoma” for a benign mixed tumor occurring in various organs including the lung. He considered these lesions not to be true tumors, but rather tumor-like structures composed of normal tissue of the organ arranged in an abnormal way or developed abnormally and showing varying degrees of differentiation of the individual tissues. The term “chondroma” still appears in articles describing lesions such as these and should probably be discarded in favor of the more descriptive and accurate term “hamartoma.” Bragg and Levine found that about 100 pulmonary hamartomata have been reported mostly under the name of “chondroma”. Of these, 75 per cent were discovered incidentally at autopsy while the remainder were surgical findings. Less than 10 per cent, however, were correctly diagnosed pre-operatively.

It might be assumed that the tumor is a rare one from the small number of cases reported. That this is not so is confirmed by McDonald, Harrington and Clagett who reported 23 cases from the Mayo clinic of which 20 were found incidentally at 7,982 necropsies and three were diagnosed correctly pre-operatively. This represents an incidence of 1 in 400 autopsies. Bragg and Levine feel that the incidence of this tumor is greater than that of bronchial adenomata and is second only to bronchogenic carcinoma. Bronchial adenomata are more rapid in their growth, occur in larger bronchi and cause symptoms of obstruction and bleeding while hamartomata, on the other hand, are slow growing, usually sub-pleural and are not likely to produce symptoms leading to their discovery.

**Figure 4**: Photomicrograph showing well differentiated epidermoid carcinoma of lung.—**Figure 5**: Photomicrograph showing benign hamartoma.
Hamartomata are found three or four times as frequently in males as in females and have occurred in all ages from nine to 87 years. The distribution is roughly proportional to the relative size of the lobes, so that the lower lobe of the right lung is the most common location. They are most often solitary, the nodules ranging in size from a few mm. to 9 cm., the majority ranging from 0.5 to 3.0 cm. Simon and Ballon⁸ report an 11 by 8 cm. hamartoma with obstructive symptoms causing pneumonia, bronchiectasis, brain abscess and meningitis which was erroneously considered to be a bronchogenic carcinoma. Though these tumors are practically always solitary, Ewing⁹ quotes Virchow as describing multiple chondromata of the lung located at the root, in the parenchyma and on the pleura.

The gross appearance is characteristic. They are lobulated, round or spherical, sharply delineated from surrounding lung tissue and usually bound by a discrete capsule which strips easily. The cut surface is avascular, whitish, never anthracotic, with the consistency and appearance of layers of cartilage separated from other tissues. Areas of calcification or islands of bone formation may offer resistance to cutting.

The microscopic picture is also characteristic. The most prevalent tissue is cartilage arranged in irregular islands, sheets or strands or less commonly as a solid mass of cartilage. The irregular islands of cartilage are usually separated by areas of connective tissue, some of which are in various stages of mucoid degeneration and have an appearance not unlike mesenchymal tissue. Interspersed throughout, there may be epithelial or glandular tissue elements resembling that of small bronchi lined by simple cuboidal or columnar cells which may be ciliated. These glands occasionally produce mucous pockets. Fibrous tissue, smooth muscle and bone are occasionally present and it is not unusual to find scattered groups of mature fat cells, especially at the periphery (Figure 4).

Diagnosis by roentgenogram is not always possible but should be made more often than it has been in the past. The appearance of a solitary sharply demarcated round or spherical tissue with lobulated margin surrounded by normal lung tissue usually located peripherally or sub-pleurally and occasionally showing patchy areas of calcification on tomogram with areas of lesser density or fuzziness at the periphery should be suggestive of a hamartoma.

Since malignant change in a hamartoma is rare, the importance lies in differentiating this from other dangerous lesions. Of these, bronchogenic carcinoma is most important to rule out. The latter are faster growing and if calcification is present, bronchogenic carcinoma is unlikely. A solitary pulmonary metastasis may be impossible to differentiate from hamartoma without calcification, as in the case presented. Since solitary metastatic lesions are now resected in selected cases,¹⁰,¹¹ thoracotomy and frozen section may decide between simple enucleation and radical lobectomy or pneumonectomy. Bronchial adenoma may occasionally resemble hamartoma, but symptoms of bleeding, obstruction, hilar location and bronchoscopy should ensure diagnosis. Pulmonary “tuberculomata” may be indistinguishable from hamartomata, especially since areas of calcification may
be seen in both. When a history of tuberculosis is not obtainable, the
presence of a peri-tuberculomatous inflammatory reaction may present a
fuzziness or haziness so that they may occasionally be indistinguishable
from hamartomata. As more tuberculomata are resected many additional
hamartomata will be discovered.

ADDENDUM

The patient expired on August 24, 1952. Autopsy revealed metastases to
tracheal lymph nodes and liver with invasion of right diaphragm. Bronchial
stump showed no carcinoma. An incidental finding was a thyroid adenoma.

SUMMARY

A case is reported of primary bronchogenic carcinoma with co-existing
hamartoma of the same lung in a 71 year old male. Pre-operatively, the
benign lesion was thought to be an ipsilateral metastasis and pneumonec-
tomy was successfully performed. Fortunately, the two tumors were in the
same lung. Otherwise, the lesions would have been considered inoperable.
The possibility of multiplicity of tumors, benign or malignant, has been
stressed.

RESUMEN

Se refiere un caso de carcinoma bronquiogénico primario con hamartoma
coeexistente en el mismo lado pulmonar en un hombre de 71 años. Antes de
la operación la lesión benigna se creyó que era una metástasis ipsilateral
y la neumonectomía fué realizada con éxito. Afortunadamente los dos tu-
mares estaban en el mismo pulmón pues de otra manera se hubiera consi-
derado un caso inoperable. Se reclama la posibilidad de la existencia de
tumores múltiples benignos o malignos.

RESUME

Les auteurs rapportent l'observation d'un homme de 71 ans, atteint d'un
cancer primitif des bronches associé à un hamartome du même poumon.
Avant l'intervention, la tumeur bénigne fut considérée comme une méta-
tase et l'on pratiqua avec succès une pneumonectomie. Il est heureux que
les deux tumeurs se trouvaient dans le même oumon. Dans l'autre cas, les
lésions auraient été considérées comme inopérables. Les auteurs insistent
sur la coexistence possible de tumeurs multiples, les unes bénignes, les
autres malignes.

REFERENCES
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