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Recurring pulmonary hamartomas: cause for concern?

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Recurring Pulmonary Hamartomas: Cause for Concern?

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Abstract

We report the case of a well-controlled female asthmatic who developed 'multiple pulmonary hamartomas' on three separate occasions over a period of 25 years that necessitated surgical resection. To our knowledge, this is the first report of recurrent hamartomas in a single individual necessitating multiple thoracotomies.

Case Report

A twenty-four year old female with asthma was referred for further evaluation with an incidental detection of a pulmonary opacity within the left lung field on routine chest radiography. She was a smoker of six pack years. Computed tomography (CT) of the chest showed a pulmonary nodule. Excision biopsy showed a benign 'pulmonary hamartoma'. She re-presented five years later, with multiple opacities on chest x-ray, and CT revealing seven calcified pulmonary nodules in the left lower lobe suggestive of a recurrence of 'pulmonary hamartomas'. No endobronchial lesion was seen at bronchoscopy. A thoracotomy and excision of the nodules confirmed benign 'hamartomas'. These lesions were confirmed as pulmonary hamartomas pathologically owing to the presence of bronchial glands, fibromyxoid stroma and cartilage (Figure 1). In view of the recurrence of the 'hamartomas', close radiological surveillance was pursued. Ten years after the second 'hamartoma' resection, radiology revealed recurrence of further nodules in even greater number (Figure 2).

She now reported dyspnoea on steep inclines and also experienced transient left sided chest pain. Her asthmatic symptoms were now well controlled with no cough or wheeze. She did describe approximately four lower respiratory tract infections annually that necessitated antibiotic treatment and only infrequently necessitated a pulse course of oral steroids. Physical examination and baseline laboratory testing including full blood count, renal and liver function and corrected calcium were unremarkable. Echocardiography and exercise stress testing were normal. CT thorax at this time illustrated four heavily calcified hamartomas and two new hamartomas within the left lung field (Figure 2).

Pulmonary function testing revealed mild obstruction with normal peak expiratory flow rates, static volume and diffusion capacity. Given the repeated recurrences of multiple parenchymal pulmonary hamartomas despite multiple resections, we ask the question 'is there cause for concern?' Referral for a thoracic surgical opinion is currently being pursued with a view to further resection.

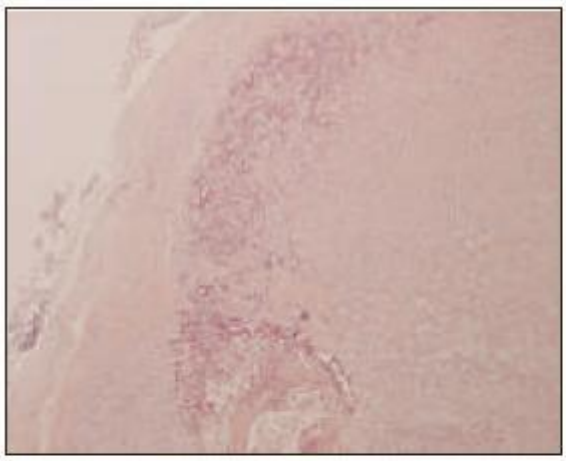

Figure 1
Haematoxylin and eosin (H & E) staining of a typical hamartoma. Note the presence of bronchial glands, fibromyxoid stroma and cartilage.

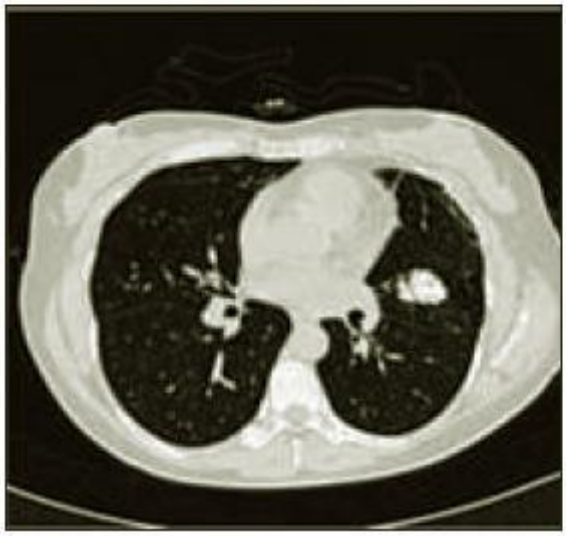

Figure 2
CT thorax demonstrating left upper and mid-zone pulmonary hamartomas.

Discussion

Pulmonary hamartomas remain the most common benign lung tumour with incidences from necropsy studies ranging between 0.025% and 0.32%.[1,2] Most are asymptomatic at diagnosis, and the hamartoma is usually detected incidentally by chest radiography. Most pulmonary hamartomas present as solitary asymptomatic peripheral nodules (intrapulmonary hamartomas). Endobronchial hamartomas are less frequent with a reported incidence of 1.4% in large published series.[3] The latter may cause airway obstruction, atelectasis and recurrent pneumonia. The term 'hamartoma' first introduced by Albrecht in 1904 was used to describe lesions containing normal organ components in an abnormal mixture. This was used to imply their benign nature with minimal malignant potential[4]. A pulmonary hamartoma may consist of fat, epithelial or fibrous tissue and cartilage, but with disorganized growth. The largest published series of 215 cases over a 17-year period illustrated that whilst the lesion is benign and most commonly detected asymptomatically in older men, a substantial number of patients had concurrent neoplasms[3].

Importantly, no evidence was found of malignant transformation of the hamartoma. Some argue that hamartomas represent a true neoplasm rather than a developmental abnormality. Cytogenetic studies have identified an abnormal karyotype in some with chromosomal bands of recombination[5]. There are however, only sporadic reported cases of malignant transformation within pulmonary hamartomas[6-8]. Based on current literature, the relationship between hamartomas and a synchronous malignancy remains a subject of ongoing debate. Several authors, including Gjevre et al[3], have reported such cases, however their significance remain unclear. Our case is unique as it does not fit the typical age or gender profile of pulmonary hamartomas at presentation and is the first reported case, to our knowledge, to necessitate recurrent surgical resections over two decades.

It is however crucial to differentiate our case of recurrent pulmonary hamartomas from pulmonary chondromas, as the latter have been documented in association with Carney's triad. This condition, which describes the coexistence of several neoplasms, including some malignant, is also predominantly seen in young women, and is frequently asymptomatic[9]. In summary, we report the case of an asthmatic female who developed multiple episodes of recurrent pulmonary hamartomas over a 25-year period necessitating multiple surgical resections hence 'a cause for concern' owing to its recurrent and

persistent nature.

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