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Multiple recurrent intrapulmonary and endobronchial mesenchymomas (hamartomas)

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Abstract

A patient is described with multiple, benign, chondromatous intrapulmonary and endobronchial mesenchymomas of the lung, which recurred after resection on two occasions over a period of 30 years. In such a patient presenting at a young age or with a history of previous recurrence, a wedge excision may be necessary to prevent further recurrence.

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Mesenchymomas of the lung (previously called hamartomas) are benign neoplasms consisting of multiple types of connective tissue and containing epithelial inclusions.[1] They may be either intrapulmonary (parenchymal) or endobronchial (3–19%),[1–4] and usually present in late middle age.[2] We report a patient who developed multiple recurrent intrapulmonary mesenchymomas over a period of 10 years, and who later developed multiple endobronchial mesenchymomas 30 years after the first resection of an intrapulmonary tumour.

Case report

At the age of 24 a calcified shadow was noted in the patient's left upper lobe or lingula on a chest radiograph. Three years later the lesion had enlarged and the patient subsequently underwent thoracotomy where a tumour was shelled out (histology not available). Four years later the patient had a small haemoptysis, and a chest radiograph showed a new calcified lesion anterior to the left main bronchus. A further four years later the patient was noted to have several chest shadows, including one at the left hilum. Bronchoscopy showed no endobronchial lesions. Thoracotomy was performed via the old wound and a large tumour was removed from the anterior segment of the left upper lobe, as well as many small tumours from the anterior edge of the left lower lobe. A further mass was palpable in the left hilum which was found to be related to the left main bronchus and this was also removed. Histology showed simple "hamartomas" with no evidence of malignancy.

At the age of 56 the patient presented with chest discomfort and chest radiography showed a calcified left hilar mass (fig 1). Fibreoptic bronchoscopy showed multiple small endobronchial mesenchymomas in the left upper lobe bronchus. Three years later the patient had a severe episode of bronchitis. Repeat bronchoscopy showed increased size of the mesenchymomas, although the bronchus was still patent. Six months later, however, a new shadow was seen on the chest radiograph, close to the previously noted mesenchymoma. Computed tomography showed this to be a soft tissue mass close to the pericardium. She again underwent thoracotomy. A firm calcified lobulated mass up to 5 cm in diameter was found occupying most of the inferior segment of the lingula, with the distal part of the bronchus containing pus. Histology of the tumour, which was shelled out intact, confirmed benign "hamartoma" with cartilage (some of which was calcified), bone, and respiratory epithelium with mucous glands. The margin of the specimen was formed by cartilaginous components with no surrounding rim of lung tissue (fig 2). Numerous other pieces of hard bony lobulated tissue were also removed from the left upper lobe.

Discussion

This patient is unusual because she developed repeated recurrences of parenchymal mesenchymomas after resection and because she also later developed coexistent endobronchial tumours. Bronchoscopy was not performed at the time of the first resection, and the rigid bronchoscopy performed at the second operation may have missed tumours in the left upper lobe bronchus.[2] It is unlikely that the endobronchial lesions were the result of "seeding" during the previous removal of intrapulmonary lesions since these had apparently been "shelled out" intact.

The presence of multiple parenchymal mesenchymomas is very rare, and parenchymal and endobronchial lesions coexist only

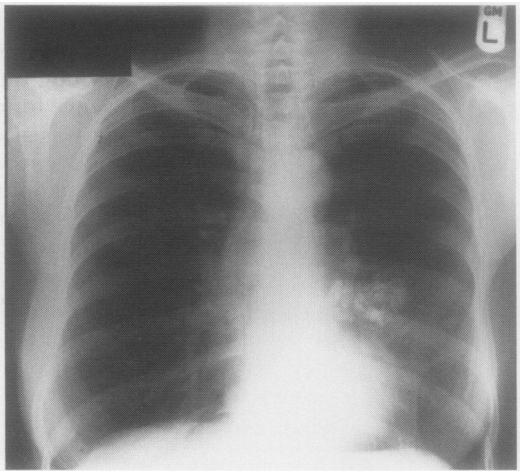

Figure 1 Chest radiograph showing calcified masses at the left hilum and left mid zone.

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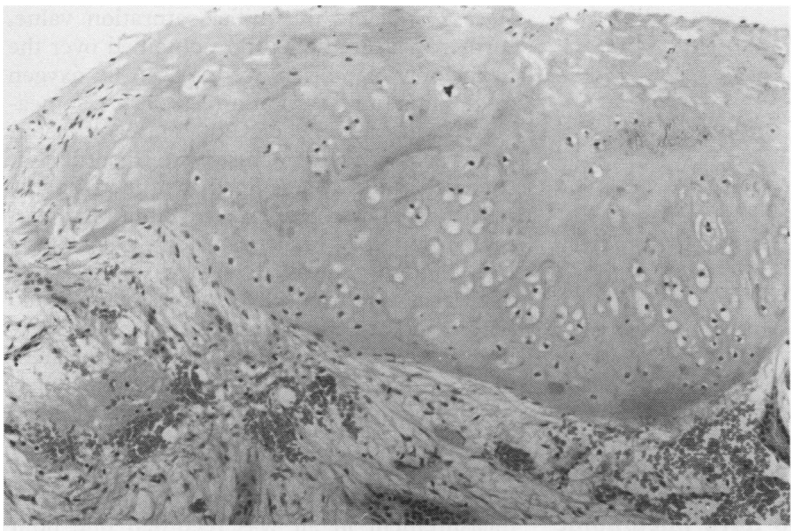

Figure 2 High power view of excision margin (top) showing lack of rim of pulmonary parenchyma. The margin is formed by cartilaginous components. Haematoxylin and eosin.

very occasionally. One report of 154 patients described four patients with two synchronous intrapulmonary lesions.[2] Another report of 33 patients found one patient with coexistent parenchymal and endobronchial tumours, and one patient with multiple parenchymal tumours.[4] Both reports found that, as with our patient, the tumours were histologically similar, supporting the view expressed by Bateson[1 3 5] that the only difference between intrapulmonary and endobronchial chondromatous mesenchymomas was their site of origin (large or small bronchi) and the direction of their growth (towards or away from the lumen of the bronchus of origin).

Recurrence of intrapulmonary mesenchymomas after section is also very rare, although a few cases have been reported.[5 6] In one study, follow up of 138 patients with parenchymal mesenchymomas found two patients in whom tumours had recurred in the same pulmonary segment 10 and 12 years later respectively.[2]

In the study of 142 patients with parenchymal tumours by Van den Bosch and coworkers, the boundary between tumour and lung was often indistinct or papillary as a result of outgrowths of fibroblasts into alveolar walls. Parenchymal tumours with a predominantly chondroid structure had a more solid appearance with a sharp border and few epithelial inclusions, although in the two patients with a recurrence, chondroid tissue was predominant in both. Despite this, the surgical technique of "shelling out" mesenchymomas appears to be sufficient to prevent recurrence in most patients, even though excision is often incomplete histologically. In the very occasional patient, such as those presenting at a young age or in those with a previous recurrence, a wedge excision with a narrow margin of normal lung may be preferable.

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