Multiple hamartomata of the lung

M. A. MADANI, C. S. DAFOE, AND C. A. ROSS

Division of Thoracic and Cardiovascular Surgery, University of Alberta, Edmonton, Alberta, Canada

Hamartoma of the lung is a rare, benign tumour. In 1963, Blair and McElvein found only 225 reported operative cases in the literature. While this tumour as a single nodule is rare, multiple hamartomata of the lung are very rare; there have been only three cases reported in the world literature (Logan, Rohde, Abbott, and Meltzer, 1965). It is the purpose of this paper to present another case of multiple hamartomata of the lung.

CASE REPORT

A.T., a 50-year-old white woman, was admitted to Aberhart Memorial Sanatorium for investigation of

spherical densities noted in radiographs of her chest. These had first been seen 11 years previously. Since that time she had had annual chest radiographs. The most recent film showed an appreciable increase in the size of the lesions.

Her past history consisted of a respiratory infection in 1939, at which time a chest radiograph disclosed a cavity in the left upper lobe (Fig. 1), and sputum examinations were positive for acid-fast bacilli. She was admitted to a sanatorium in Saskatchewan and later underwent a left thoracoplasty. In 1944 she was classed as having arrested tuberculosis, and she resumed her personal and social activities. She was checked annually at the sanatorium, and no evidence of reactivation of the pulmonary lesion was found.

FIG. 1. (1939) Cavity measuring 3.5 cm. in diameter located in the first interspace, left lung field.

FIG. 2. (1954) *Densities in the left lung field as follows : below the anterior end of the second rib, 0.7 cm.; overlying the fourth rib anteriorly, midzone, 2.3 cm.; just above the fifth rib anteriorly, midzone, 0.9 cm.*

FIG. 3. (1958) *Densities in the left lung field as follows : below the anterior end of the second rib, 0.9 cm.; overlying the fourth rib anteriorly, midzone, 2.4 cm.; just above the fifth rib anteriorly, midzone, 0.9 cm.; above the cardiophrenic angle, 0.5 cm.*

FIG. 4. (1961) Densities in the left lung field as follows : below the anterior end of the second rib, 0.9 cm.; overlying the fourth rib anteriorly, midzone, 2.5 cm.; just above the fifth rib anteriorly, midzone, 1 cm.; above the cardiophrenic angle, 1 cm.; above the diaphragm centrally on the right, 0.5 cm.; over the sixth rib posteriorly, 0.8 cm.

FIG. 5. (1965) Densities in the left lung field as follows : below the anterior end of the second rib, 1 cm.; mid-zone in the upper border of the second rib, 1 cm.; overlying the fourth rib anteriorly, midzone, 3 cm.; above the fifth rib, 1.5 cm.; above diaphragm centrally, 1 cm.; above the cardiophrenic angle, 1.5 cm. Densities on the right side as follows : over the sixth rib posteriorly, 1 cm.

FIG. 6. Multiple hamartomata of the lung: (a) low and (b) high magnifications.

(a)

(b)

In 1954 she suffered an acute upper gastrointestinal haemorrhage and was admitted to University Hospital, Edmonton. At that time the chest radiograph showed multiple densities in the right lung field (Fig. 2). On the basis of a past history of tuberculosis, these densities were interpreted as tuberculoma and, as far as treatment was concerned, they were disregarded. She continued to have annual chest radiographs. In 1958 and in 1961, attention was paid to the gradual increase in size of these densities and also to the appearance of additional nodules (Figs 3 and 4). On each occasion it was the opinion of the internist in charge of this patient that a programme of frequent careful re-examination was all that was necessary.

In July 1965 the increase in size of the nodules was significant (Fig. 5). The patient was admitted to Aberhart Memorial Sanatorium for investigation and reassessment of these densities. She had remained asymptomatic. Examinations for tubercle bacilli were negative; the histoplasmine and Casoni tests were also negative. Investigation of the gastro-intestinal and urinary tracts disclosed no lesion. In view of the significant enlargement of the lesions in the chest, a thoracotomy was advised. She was operated on in August 1965, and exploration of the right lung revealed the nodules to be in the periphery of the lung. Three of the most superficial lesions were removed; frozen section excluded malignancy. The pathological report was as follows:

GROSS APPEARANCE There were three nodular specimens, measuring $3 \times 1.5 \times 1$ cm., $2 \times 1 \times 1$ cm., and $1.2 \times 0.8 \times 0.5$ cm., each having a slightly irregular surface composed of brown and white tissue. The cut surface revealed whitish tissue in which there were several small cysts.

MICROSCOPIC APPEARANCE All specimens showed the same histological features, which consisted of interlacing bundles of smooth muscle, within which there were vascular channels. There were occasional well-defined vascular channels and numerous cystic spaces of varying size which were lined by low cuboidal to flattened epithelium. The periphery of each lesion was well demarcated from the adjacent pulmonary parenchyma, some of the adjacent alveolar spaces of which exhibited foetalization of their linings. No fatty tissue or cartilage was present, but the histological features were otherwise compatible with benign hamartoma (Figs 6a and b).

DISCUSSION

From a review of the literature, we found that multiple hamartomata of the lung are rare. Deussig was the first to report such a case in his thesis in 1912 (cited by Logan). We had no opportunity to review this case as the thesis was burned in the first world war. The second and third cases of multiple hamartomata of the lung were reported by Keers and Smith (1960) and Logan *et al.* (1965) respectively.

In reviewing the pathological reports of the patients of Keers and Smith, and Logan *et al.*, as well as of the present case, cartilaginous elements have been absent, whereas the single hamartoma very frequently contains cartilage. This might be of pathological significance and may have some connexion with the multiplicity of the tumours.

Since multiple hamartomata will usually involve both lungs and multiple lobes, the question of operative intervention is open to discussion.

In view of the fact that the world literature shows no evidence that these lesions become malignant, it seems logical that, after the diagnosis has been confirmed by thoracotomy, the lesions on the contralateral side may be safely left *in situ* under continued regular radiological observation. It seems preferable to operate on the side showing the greatest pathology and to remove as many lesions as is technically feasible, with the preservation of as much lung tissue as possible.

SUMMARY

A case of multiple hamartomata of the lung is presented. This is the fourth case reported in the world literature.

None of these lesions had cartilage as a component of the tumours.

REFERENCES

- Blair, T. C., and McElvein, R. B. (1963). Hamartoma of the lung. *Dis. Chest*, **44**, 296.
- Deussig, R. (1912). Multiple primary myoma of the lung. Thesis 1912, University of Munich. Not available (personal communication with the University 4/10/1965).
- Keers, R. Y., and Smith, F. A. (1960). A case of multiple "hamartomata" of unusual type. *Brit. J. Dis. Chest*, **54**, 349.
- Logan, W. D., Rohde, F. C., Abbott, O. A., and Meltzer, H. D. (1965). Multiple pulmonary fibroelastomyomatous hamartoma. *Amer. Rev. resp. Dis.*, **91**, 101.