An unusual presentation of sarcoidosis—spontaneous haemopneumothorax

STEPHANIE A. GOMM*
M.B., Ch.B., M.R.C.P.

North Manchester General Hospital, Crumpsall, Manchester 8

Summary

Pneumothorax is a very rare complication of early stage sarcoidosis. A young man is reported with a left sided spontaneous haemopneumothorax and a right apical pneumothorax in association with early sarcoidosis.

KEY WORDS: haemopneumothorax, sarcoidosis.

Introduction

Pneumothorax is reported as occurring in 2-4% of patients with sarcoidosis but is usually observed as a complication of late stage fibrotic and bullous disease and has rarely been described in early sarcoidosis. A patient, previously asymptomatic, is reported who presented with a left sided haemopneumothorax and a right apical pneumothorax. A subsequent lung biopsy showed a histological diagnosis of sarcoidosis. The occurrence of haemopneumothorax complicating bilateral pneumothoraces in early stage sarcoidosis has not previously been reported.

Case report

In February 1981, a 29-year-old barman was admitted with a 3-day history of left sided pleuritic chest pain. Twenty-four hours before admission he complained of breathlessness at rest, sweating and light-headedness on standing. There was no previous history of fever, productive cough, haemoptysis or dyspnoea. He was a non-smoker. In 1967 he had had a polya-gastrectomy for a perforated duodenal ulcer.

On examination he was a thin man, clinically anaemic and afebrile. There were signs of a large left pleural effusion. A chest radiograph (Fig. 1), showed a right apical pneumothorax and a large left pneumothorax with a fluid level to the seventh rib anteriorly. The haemoglobin was 8.7 g/dl.

Four and one-half litres of heavily blood-stained fluid was drained from the left pleural cavity and the

FIG. 1. Chest radiograph showing a right apical pneumothorax and a left sided haemopneumothorax.

left lung re-expanded. Twenty-four hours later further bleeding occurred. An explorative left-sided thoracotomy was performed, at which 200 ml of organized blood clot was removed and the lung was noted to be covered in a fibrinous material. The only source of bleeding was an area of capillary oozing near the thoracic inlet. A number of small nodules were noted on the surface of the lung, one of which

*Present address: Department of Chest Medicine, Wythenshawe Hospital, Southmoor Road, Manchester 23.

was excised. His postoperative recovery was uneventful.

The histology of the lung tissue showed multiple granulomata composed of epithelioid cells and multinucleated giant cells in the sub-pleural connective tissue and in the interstitial tissue of the underlying lung (Fig. 2) There was no evidence of caseation and several of the granulomata were healing by fibrosis. Inclusion bodies of the Schaumann conchoidal type were seen. Cultures for acid-fast bacilli and fungi were negative.

FIG. 2. Lung biopsy showing non-caseating granulomata.

Subsequent investigations showed normal biochemistry. A Mantoux test was negative at 1/1,000. Lung function tests 3 months after his thoracotomy showed a moderately severe restrictive ventilatory defect. The transfer factor was similarly impaired. Serum angiotensin converting enzyme was 105 iu/ml (normal range 15–55). Six months later he had developed purplish skin nodules on the surface of the thoracotomy scar and subsequent biopsy showed typical non-caseating granulomata. His chest radiograph showed a resection of the left sixth rib anteriorly with obliteration of the left costo-phrenic angle, bilateral

hilar and right paratracheal lymphadenopathy was present but there was no evidence of pulmonary infiltration.

Discussion

The association between pneumothorax and early sarcoidosis could be causal or coincidental. Riley (1950) first reported pneumothorax as a complication of sarcoidosis in 2 of a series of 52 patients and proposed that the pneumothorax was directly related to either a rupture of a subpleural bleb or necrosis of a subpleural granuloma. Scadding (1967) reported 5 cases of sarcoidosis complicated by pneumothorax. Three patients were young men in whom he felt the pneumothorax was coincidental, but the other 2 were black women with late stage bullous and fibrotic disease with recurrent pneumothoraces. Sharma (1977) reviewed 180 cases of sarcoidosis and found that pneumothorax developed in 5 patients with interstitial lung infiltration, 2 of whom had non-caseating granuloma involving the pleura at thoracotomy. Thus in certain cases pneumothorax may be directly caused by necrosis of a subpleural granuloma.

In this young man a diagnosis of early sarcoidosis was made on the histological basis of sub-pleural and interstitial lung non-caseating granuloma in which no acid-fast bacilli or fungi were seen. Subsequent biopsy of a skin nodule showed typical non-caseating granulomata. A tuberculin test was negative and the serum angiotensin converting enzyme was elevated. Pulmonary function tests revealed a restrictive defect. A chest radiograph 6 months postoperatively showed bilateral hilar and right paratracheal lymphadenopathy. Two years later he is asymptomatic and his chest radiograph remains unchanged. Thus the development of bilateral pneumothoraces complicated by a left sided haemopneumothorax represents an early feature of sarcoidosis. Ross and Empey (1983) reported a case of bilateral pneumothoraces in early sarcoidosis. However, the association of sarcoidosis and bilateral pneumothoraces with the unusual complication of a haemopneumothorax without radiographical evidence of pulmonary infiltration has not previously been described.

Acknowledgments

I wish to thank Dr B. A. Enoch, Dr M. O'Connor, Mr M. D. Fitzgerald, Dr J. S. Whittaker and Dr Dekretzer for their cooperation in the preparation of this case report, and Miss M. Culliney for secretarial assistance.

References

- RILEY, E.A. (1950) Boeck's sarcoid: a review based upon a clinical study of 52 cases. *American Review of Tuberculosis and Pulmonary Diseases*, **62**, 231.

ROSS, R.J.M. & EMPEY, D.W. (1983) Bilateral spontaneous pneumothorax in sarcoidosis. *Postgraduate Medical Journal*, **59**, 106.
SCADDING, J.G. (1967) *Sarcoidosis*, 1st edn, p. 136. Eyre and Spotiswoode, London.

SHARMA, O.P. (1977) Sarcoidosis: unusual pulmonary manifestations. *Postgraduate Medicine*, **61**, 67.

(Accepted 4 August 1983)