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The Lymphographic Findings in a Case of Lymphocytic Lymphoma Presenting as Recurrent Chylothorax

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Summary

A case of lymphocytic lymphoma is presented. The unusual mode of presentation as a result of bilateral chylothorax is described. The value of lymphography in establishing the site of lymph leakage and in the establishment of the underlying diagnosis is demonstrated and discussed.

Introduction

The occurrence of pleural effusions in patients suffering from one of the lymphomas is a common complication, usually occuring in the late stages of the disease.

Dyspnoea, as a result of pleural effusions, as a presenting symptom in the lymphomas is unusual but *Ngan* and *James* quote three cases reported by *Ch'en* et al. (1966) where dyspnoea as a result of pleural effusions was the presenting symptom. A small proportion of the pleural effusions occurring in patients with lymphoma are chylous, whereas the majority are clear effusions presumably arising as a result of direct involvement of the pleura by the disease process, or possibly by the obstruction of the pulmonary veins by enlarged nodes in the hilum and mediastinum. In addition, hypo-proteinaemia appears to be of significance in these cases (*Ch'en* et al. 1966). Obstruction to lymph flow by lymph nodes involved with lymphoma may occur, although this is not a finding particularly associated with lymphomatous glands. Where obstruction to the flow of the lymph does occur in the mediastinum a chylous effusion may result, and may be precipitated by trauma (*White* and *Urquhart* 1966).

Lymphography is widely used as one of the routine staging procedures in patients presenting with lymphomas and in a case presenting with chylous effusions this examination could be reasonably expected to yield useful information with respect to the source of lymph leakage (*Fuchs* et al. 1969).

Case History

The patient, a 66 year old lady originally presented in December, 1974 complaining principally of a curious sensation in her chest which made her feel that she must "hold her breath". This symptom had occurred previously in the preceeding two years, lasting over the winter months and usually commencing in October. On the occasion of her presentation this symptom had been noticeably more severe than previously. There was no history of tightness in the chest or wheezing, but there was a history of some exertional dyspnoea although this was not of any great severity. She had noted attacks of tachycardia occasionally accompanying these other symptoms. On examination the only abnormality was a diminution of breath sounds and the percussion note at the left base. Her chest X-ray at this time (Fig. 1) shows bilateral pleural effusions, that on the left being larger than that on the right, and no obvious lesion in the lung fields or mediastinum. The peripheral blood picture at the time of presentation was normal. Following these findings, the patient was admitted to hospital for investigation of her pleural effusions and was then complaining of considerable dyspnoea. It was noted that, on admission to hospital, approximately one month after the original consultation, the right pleural effusion was now larger than the left. Pleural tap was carried out and yielded 300 ml of milky fluid. A pleural biopsy was obtained at the same time. This showed no evidence of tumour or tuberculosis. Mantoux test was negative. The pleural fluid was found

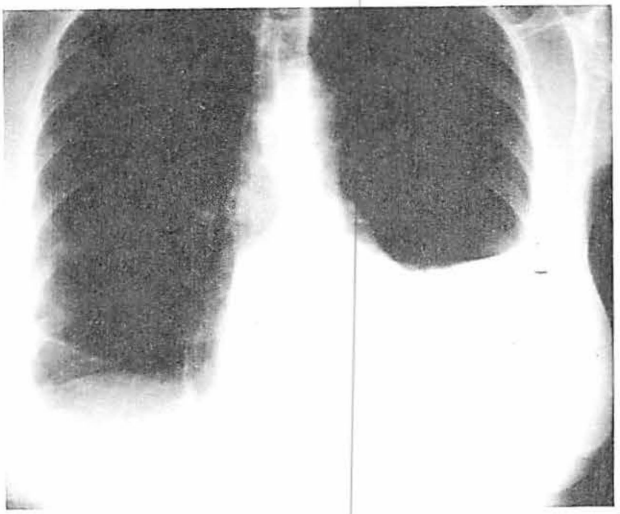

Fig. 1: Chest X-ray at original presentation showing large left pleural effusion and small right effusion.

to contain many lymphocytes and some mesothelial cells and its triglyceride content was 650 mg percent and the protein content was 4.5 g percent, confirming leakage of lymph into the pleural cavity. While in hospital, the effusions were found to regress and the patient was discharged to be followed up as an out-patient with no definite cause for the chylothorax having been established. During the subsequent three month the patient again became increasingly dyspnoeic and also began to lose weight. She was readmitted to hospital in March as a result of her increasing breathlessness. She was then found to have a large right sided effusion, and a trace of fluid on the left (Fig. 2). Lymphangiography was carried out on April 15, 1975 (Figs. 3, 4 and 5) employing standard technique with an injected volume of 5 ml ultra-fluid Lipiodol® on each side. This revealed normal glands in the pelvis but enlargement of the para-aortic glands with numerous small fillings defects and irregular margins (Fig. 4). These features were considered to be typical of nodular sclerosing Hodgkin's disease, though not necessarily diagnostic of this condition as

Fig. 2: Chest X-ray on admission to hospital shows almost complete clearance of the left pleural effusion but enlargement of the effusion on the right.

Fig. 3: Lateral view of chest showing filling of terminal portion of thoracic duct.

many other conditions can give rise to similar radiological appearances. In addition a number of other very interesting features were noted. Firstly, the presence of contrast in lymphatic vessels on the 24 hour films (Fig. 4) was suggestive of obstruction to the flow of lymph proximally as was the abnormal filling of a number of abdominal lymphatic vessels not usually seen (Fig. 6). Obstruction if present was clearly not complete as contrast was seen in the terminal portion of the thoracic duct (Fig. 3). In the localised view of the region of the right cardiophrenic angle (Fig. 4) a lymphatic channel was seen running laterally and inferiorly into the right pleural cavity. At its distal end a few free-lying droplets of Lipiodol® were identified within the pleural effusion. On the lateral view (Fig. 5) Lipiodol® can be seen lying free on the superior aspect of the right dome of the diaphragm. Biopsy of lymph node showed typical changes of a well differentiated nodular lymphocytic lymphoma and the patient was

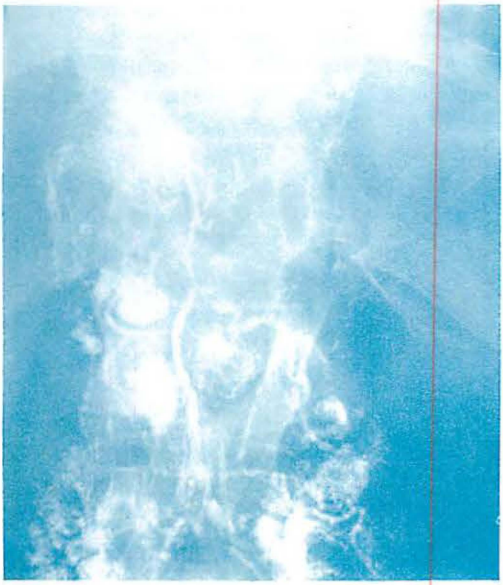

Fig. 4: A.P. view of right cardiophrenic angle showing abnormal glands and filling of a basal pleural lymphatic running obliquely into the right pleural cavity and ending in a few free-lying droplets of Lipidol. The nodes are enlarged, containing numerous filling defects and show marginal irregularities.

Fig. 5: Lateral view of the same area showing the basal pleural lymphatic running downwards and posteriorly and free Lipiodol on the superior aspect of the diaphragm.

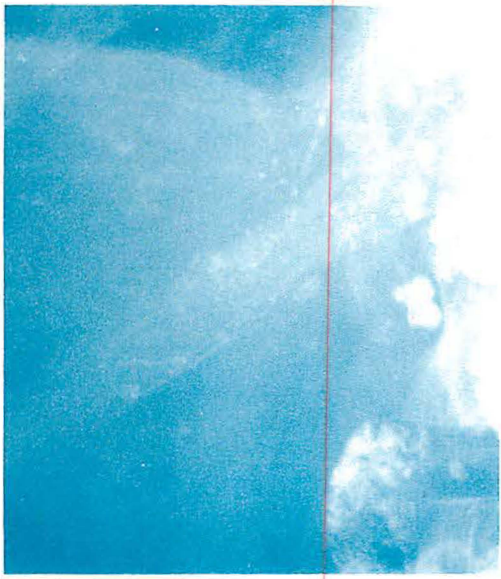

Fig. 6: Immediate post-injection phase. The radiograph shows filling of numerous lymphatic vessels which are not normally seen, good filling of the thoracic duct and evidence of gland enlargement in the para-aortic chains.

subsequently treated by radiotherapy and chemotherapy, but, during, chemotherapy developed a gram-negative septicaemia from which she subsequently died. At autopsy, no residual lymphomatous disease was identified.

Discussion

In view of the greatly improved prognosis for patients suffering from lymphoma as a result of modern treatment, there is now increased emphasis on early diagnosis of the condition and accurate staging of the extent of the disease (*McDonald* 1973). At this centre, lymphography is routinely used in patients presenting with lymphoma whose clinical stage is either I, II or III at presentation. In stage IV cases, the examination is occasionally carried out in order to monitor the patient's response to treatment. All lymphoma patients who have had lymphography carried out in fact receive regular radiographic follow-up examinations for a period of at least six months. The present case was somewhat unusual in that the diagnosis of the lymphoma was suggested on the basis of the lymphogram. The staging as a result of this examination would be stage III (abnormal glands on both sides of the diaphragm, Fig. 5). There is no definite proof radiologically of involvement of either lung or pleura to place the patient in stage IV.

Chylothorax is in itself a rare condition which is most commonly seen as the result of direct surgical trauma to the lymphatics at thoracotomy (*McCormack* 1975). Indirect trauma taking the form of hyperextension injury to the dorsal spine has also been reported as giving rise to the condition (*Steiner* 1971). It may also occur as a result of congenital anomalies such as a lymphangiomatous malformation in the thoracic cavity or as a result of obstruction to the thoracic duct by metastatic deposits in the mediastinum (*Felson* 1973). In the case under discussion, although there was indirect evidence of obstruction to lymphatic flow (Fig. 4), in that contrast can be seen remaining in the lymphatic vessels 24 hours after injection and contrast can be seen filling vessels which are not normally demonstrated, total obstruction was absent as there was good demonstration of the terminal portion of the thoracic duct (Fig. 3). In a case of chylothorax or chylous ascites it could be reasonably expected that lymphography would show a site of leakage as in this case where a lymphatic channel is seen running into the chylothorax on the right side and ending abruptly in a few free-lying droplets of Lipiodol®. The characteristics of this vessel are virtually identical to those described by *Grant* and *Levin* as representing basal pleural lymphatics (*Grant* and *Levin* 1974). The appearances however do not suggest delineation of pulmonary lymphatics as described by these authors and by *White* and *Urquhart* (1966). The particular lymphatic channel concerned is seen to lie extremely far posteriorly as seen on the lateral view (Fig. 5) and in fact overlies the posterior one third of the body of D12. It is reasonable to suppose that lymphatics in this area would be particularly susceptible to rupture in the hyperextension of the spine, but as there is no history of such trauma in this case it

seems likely that the mechanism was obstruction by involvement of lymphatic glands in the mediastinum. It is also of interest to note that although lymphographically abnormal glands are demonstrated in the mediastinum, at no time was there evidence on chest radiography of mediastinal gland enlargement.

Acknowledgements

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References

Ch'en, Y.S., K.J. Chang, T.E. P'eng, C.M. LI, S.H. Ch'en: Malignant Lymphomas with special Manifestations. Chinese Medical Journal 85 (1966) 229-235

Felson, B.J.: Chest Roentgenology. Saunders, Philadelphia (1973) pp. 20-21

Fuchs, W.A., J.W. Davidson, H.W. Fischer: Recent Results in Cancer Research; Lymphography in Cancer. Springer Verlag, Heidelberg-Berlin-New York 1969, pp. 105-106

Grant, T., B. Levin: Lymphangiographic Visualisation of Pleural and Pulmonary Lymphatics in a Patient without Chylothorax. Radiology 113 (1974) 49-50

McCormack, R.J.: Personal communication (1975)

McDonald, J.S.: Diagnostic Radiology in Hodgkin's Disease. Brit. J. Hosp. Med. (1973) 437-443

Ngan, H., W.K. James: Clinical Radiology of the Lymphomas. Butterworths, London, pp. 54-56

Steiner, W.R.M.: Roentgenographic Demonstration of Intra-Pulmonary and Pleural Lymphatics during Lymphangiography. Radiology 100 (1971) 533-539

White, W.F., W. Urquhart: The Demonstration of Pulmonary Lymphatics by Lymphography in a Patient with Chylothorax. Clin. Radiol. 17 (1966) 92-94

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