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Bilateral Chylothorax – Complication in Malignancy

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Summary

Bilateral chylothorax is a rare condition caused by traumas and disorders with perforation or destruction of main lymphatics in the thorax leading to lymph leakage. Two patients with bilateral chylothorax are presented one of whom was successfully operated on. Etiology, diagnostics and therapy of this condition are discussed.

Leakage of lymph to the pleura, chylothorax, is a very uncommon form of pleural effusion but merits attention on account of the diagnostic and therapeutic peculiarities. The condition is generally elicited by trauma, injuries in the lower part of the thorax usually leading to right-sided chylothorax, injuries in the upper part to left-sided (1). Another fairly common cause is tumour, the effusion arising from obstruction and/or erosion of the thoracic duct (2). Occasional cases have been reported of ruptured cysts or lymphangioma in the thoracic duct, as well as specific infections such as tuberculosis and filariasis (3). As a rule the effusion is unilateral. Only a few cases have been published with bilateral chylothorax.

Several surveys have been made of case reports. Reviewing the literature up to the mid-1950s, *Nix et al.* (4) found 479 cases of chylothorax but only 155 satisfied their criteria; of these, 32 were combined with chyloperitoneum. In the period 1945-65 *Kuntz* (5) found 300 cases, 19 of them bilateral. Six of them had been caused by occlusion of the thoracic duct as a result of neurinoma (1 case), polyserositis tuberculosa (1 case), pleural endothelioma (1 case), lymphogranulomatosis (2 cases) and reticulum-cell sarcoma (1 case); the last of these also had chyloperitoneum. The material of *Nix et al.* (4) included 8 cases of unspecified malignancy with uni- or bi-lateral chylothorax.

We have had two patients at the Serafimer Hospital with bilateral chylothorax due to tumour. One of the patients was operated on successfully but died two years later in another malignant disorder. The other died in the tumour disease after a rapid, asymptomatic course before surgical therapy could be instituted.

Case 1 A woman of 65 with a history of chronic lymphatic leukemia was admitted as an emergency case for rapidly increasing dyspnoea. Lung X-ray showed bilateral hydrothorax up to the hilus. Large amounts of a pale yellow, opalescent fluid were drained repeatedly and were found to contain 830 mg% total lipids, 3.5 g% protein and great quantities of lymphoid cells. Pleural drainage with continuous suction was installed bilaterally and yielded much chyle. After almost two weeks the drainage was removed because it was functioning poorly.

The rapid accumulation of chylus continued, necessitating first a left-sided drain which functioned well and then, when the amount of chyle showed no tendency to diminish after 3 weeks' treatment, an explorative thoracotomy, possibly with ligation of the thoracic duct. The thoracotomy was performed from the right side and the duct was dissected free in the mediastinum. Two hours before the operation the patient had received a tablespoon of maize oil per os in order to visualize the thoracic duct. This presented a whitish colour and stasis, the diameter at the widest point being 5-6 mm. An injection of 1 ml patent blue was given subcutaneously over the dorsum of each foot in order to dye the lymph in the thoracic duct via the lymph capillaries. The lymph in the duct turned faintly blue but no leakage was observed. Using two thin plastic catheters, distal as well as proximal lymphangiography of the thoracic duct was then performed with a view to detecting additional main trunks, which would also have to be ligated in order to eliminate any leakage.

It was found that the lower part of the thoracic duct and the cisterna chyli filled with contrast medium but that there was no leakage of this. The thoracic duct and a small branch towards the left side in the mediastinum were then ligated. Remarkably enough, no pathological changes with enlarged lymph nodes were observed in the mediastinum. Pleural drainage was provided before closing the thorax. Both drains could be removed after one week and repeated check-ups with pulmonary X-ray showed no further chylothorax.

Laboratory data: Hb 8.8 – 14.5 g/100 ml, platelets 200,000/ μ l, W.B.C. 130,000 – 51,200/ μ l, differential count 95% lymphocytes, ESR 97 – 10 mm, creatinine and electrolytes normal, serum protein 5.0 – 6.2 g/100 ml, urine analysis normal.

Case 2 A woman of 55 was admitted as an emergency case for cough and increasing dyspnoea. Essentially healthy hitherto. The current complaints started 3 weeks before admission, with unproductive cough and a slightly elevated temperature, which was interpreted as a respiratory infection. Increasing dyspnoea started one week before admission and forced the patient to sleep sitting up. On admission the woman was found to be severely dyspnoeic with a respiratory rate of 38 and a heavy, rattling cough; temperature 38.6°C. Some isolated lymph nodes were palpated on one side of the neck. ECG showed regular sinus tachycardia, 130/min. Pulmonary X-ray revealed large amounts of pleural fluid bilaterally. Her condition was interpreted preliminarily as infection and cardiac failure; routine treatment produced a substantial improvement. Next day about 600 ml of a yellow, cloudy, adourless fluid was drained from each pleura. No bacterial growth, no acid-resistant rods and no malignant cells were demonstrated in the fluid. It contained 6.5 g protein/100 ml and a prolific, predominantly lymphomonocytic cell flora. Fluid accumulated in both pleurae only a few days after the first thoracocenteses and this had to be repeated. During the 12 days that treatment lasted, the patient was drained of a total of 5,100 ml of fluid, divided between five occasions. Repeated pulmonary X-ray examinations showed increasing pleural fluid bilaterally but no parenchymal changes. B.-scan gave no indication of exudate in the pericardium.

It was suspected early on that the pleurae contained chyle and this was verified from the content of triglycerides: 505 mg%.

Apart from the periodical dyspnoea and cough, the patient's condition was not generally affected during the first week but after that she became more and more fatigued and died suddenly 12 days after admission in a state of abrupt unconsciousness, cyanosis and irregular cardiac activity.

Lymphangiography was attempted 48 h post mortem via the cisterna chyli. Numerous soft para- and pre-aortal lymph nodes up to the size of a date were found in the abdomen, making it difficult to detach any sizeable lymph vessels; no distinct cisterna chyli was seen. Contrast medium was injected into a number of the large lymph vessels but each time it leaked out into the abdominal cavity somewhere along their course. When three unsuccessful attempts had been made to fill the thoracic duct, the examination was abandoned. The parcel of thoracic and abdominal organs was then removed and turned upside down, whereupon the thoracic duct was located and injected with physiological saline solution in the lower posterior mediastinum. The duct filled in a cranial direction up to a tumorous growth measuring 8 x 5 x 5 cm in the upper mediastinum and invading both pleurae. The medium escaped from several holes in the parts of the tumour facing the right and the left pleura. The part of the thoracic duct leading to the left cervical veins (see Figure) above the tumour did not fill.

The autopsy revealed more than 2000 ml of a chylous fluid in each pleura, the serous coats of which displayed a scattering of greyish-white, tumours excrescences. The spool-shaped tumour mentioned above was located on a level with the hilus in the mediastinum and behind the oesophagus, compressing this as well as the trachea and right main bronchus. The mediastinum also contained a large number of enlarged lymph nodes, which like the tumour were soft, and grey to yellowish-white in section. There was nothing remarkable about the lung parenchyma or the pericardium. The abdominal cavity held 50 ml of a chylous fluid. The spleen weighed 530 g and sections presented deposits of a greyish-yellow infiltrate the size of a pea. The liver was of normal size. Numerous soft, enlarged lymph nodes with the same appearance as above were found along the aorta and vena cava as well as in the mesentery, pelvis and both sides of the neck.

Laboratory data. Hb 16.5 g/100 ml, W.B.C. 4,600/ μ l with normal differential count, ESR 4 mm, serum creatinine, electrolytes, transaminases, L alkaline phosphatases, AST, ASTA, cold agglutination, proteinbound thyroxine, blood lipids and urine analyses all normal. Repeated blood-gas analyses showed arterial hypoxaemia with pO₂ 52 – 60 mm Hg; otherwise normal.

Microscopic pathology: general reticulum- cell sarcoma.

Discussion

The presence of chylothorax is usually suspected when thoracocentesis yields a milky fluid. Chylothorax cannot be distinguished from other pleural effusions in X-ray pictures. The appearance may be mistaken for empyema. The chylous fluid is, however, odourless. A milky liquid is also associated with pseudo-chylous and chyliform effusions but in the former case there is a high content of un-preesterized cholesterol due to the decomposition of endothelial cells, erythrocytes and leucocytes. This is usually seen in chronic pulmonary and pleural disorders and develops slowly. Chyliform effusions are also seen in chronic pleural exudates and require the decomposition of fat-degenerated cells. The content of triglycerides is below 500 mg% and the protein content below 3 g%.

The criteria for chylothorax have been listed by Kuntz (5) as:

1. Macroscopically milky fluid
2. Protein content more than half the serum concentration
3. Triglycerides more than 500 mg%
4. Microscopically prolific lymphocytes

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All these criteria were met in case 2. In case 1 there were no data on triglycerides. The total amount of fat, 830 mg%, is within the normal limits for total plasma lipids and probably not of diagnostic significance by itself. The rapid accumulation of fluid in the pleurae coupled with the histological picture of lymphocytes made the diagnosis highly credible and this was confirmed by the finding of a distended thoracic duct and no further effusion after this had been ligated.

The chylothorax was bilateral in both cases. The rareness of this condition is explained by the mediastinal course of the thoracic duct, usually running to the right of the midline in the lower part and then to the left in the upper, so that bilateral leakage is likely to occur only upon injury halfway up the mediastinum. The tumour in case 2 was located centrally in the upper mediastinum (see Fig.) The bilateral chylothorax is more difficult to explain in case 1: no leakage was detected either from the injection of patent blue or during lymphangiography during the thoracotomy operation.

A thoracic duct damaged by malignancy may rupture as a result of, for instance, vomiting or a persistent cough and lead to chylothorax (6, 3). Uni- or bi-lateral chylothorax may be the first symptom of a progressive lymphoma (4, 1, 7).

It is now agreed that the most effective treatment for chylothorax- uni- or bi-lateral – is surgery with ligature of the thoracic duct in the mediastinum below the leakage (8, 9, 10, 11). Peroperative ductography is essential in order to locate the lymph vessels, as all these must be ligated if treatment is to be successful (12).

If the ligature is to have the intended effect, moreover, there must be collaterals – possibly lymphovenous anastomoses – which can take over the transportation of the thoracic-duct lymph past the site of the leak or tumour (13). The desired result was achieved in case 1 with surgical treatment. A conceivable alternative would have been to establish an anastomosis between the thoracic duct and the azygos veins.

In case 2, an operation was ruled out by the patient's general condition. There were, moreover, no collaterals in the mediastinum for transporting thoracic-duct lymph because the whole of the upper mediastinum had become overgrown by the primary tumour, which probably also had a fatal effect on the function of vital organs in this area. Lymphangiography ad modum *Kinmonth* (14) would have been valuable diagnostically but presumably could not contribute to effective therapy.

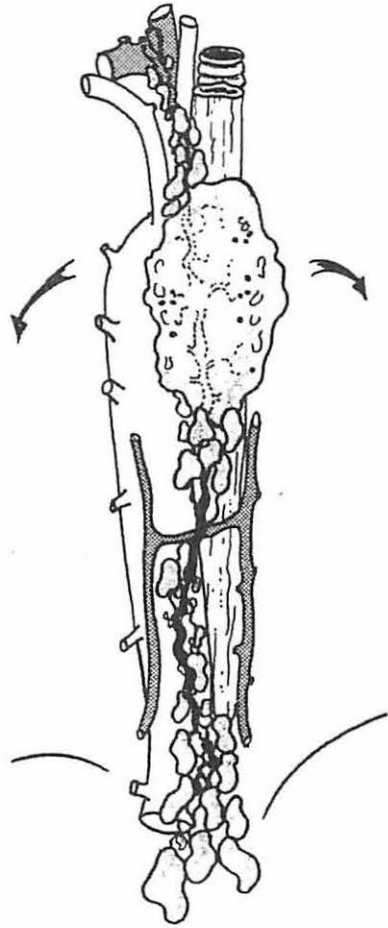


Fig. 1 Autopsy specimen (mediastinum from behind) showing lymph from the thoracic duct leaking into the right and left pleurae (black arrows) from a tumour in the upper mediastinum.

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Xeroradiography of the Lymphatics

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Summary

Lymphograms of 40 patients were made on xeroradiographic plates and conventional X-ray films. Both imaging procedures are compared in regard to their spatial and contrast resolution. Although xeroradiography offers little advantage in retroperitoneal lymphography, it essentially improves imaging of brachial lymphatic vessels and lymph nodes.

Introduction

Although xeroradiography has been widely used in X-ray examinations of breast (1-4) and bone (5-8), studies of the lymphatics by xeroradiography have not yet been reported on.

Salient features of xeroradiography in comparison to conventional X-ray film examination are

- 1) Large contrast range permitting to represent both bone and soft tissue structures at the same time

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