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CASE REPORT

The squirting bleb: image and treatment of inspiration-linked leakage of a peritoneopleural fistula
F H Krouwels, P Bresser

A 25 year old man with known chronic right sided heart failure and ascites due to a congenital heart defect presented with dyspnoea and a massive pleural effusion. Thoracoscopy revealed two diaphragmatic blebs. Changes in peritoneal and thoracic pressure during respiration resulted in periodic squirting of a ruptured bleb, illustrating preferential flow of peritoneal fluid into the thorax. The pleural effusion was successfully treated with drainage of ascitic fluid and chemical pleurodesis.

A 25 year old man was referred to our clinic with gradually increasing exertional dyspnoea. He was known to have a congenital univentricular heart for which he had had a corrective Fontan operation in 1980. He had since suffered from chronic right sided heart failure resulting in a protein losing enteropathy and ascites which was treated with diuretics and a diet of medium chain triglycerides.

On presentation he was severely dyspnoeic, had gained 7 kg in weight, and his abdomen was enlarged. Chest radiography revealed a massive left sided pleural effusion and a large amount of ascitic fluid was seen on ultrasound examination. He also had raised venous pressure and hypoalbuminaemia.

The patient was treated with tube drainage of the pleural fluid and diuretics; 4 litres of pleural fluid with characteristics of a transudate (albumin 3 g/l, lactate dehydrogenase (LDH) 34 U/l; serum levels: albumin 15 g/l, LDH 212 U/l) were removed. Because of persistent pleural fluid production of more than 0.5 l/day, an inspection thoracoscopy was performed. A tense bleb was seen on the diaphragm and another bleb was visible which was ruptured and forcefully squirted fluid into the thoracic cavity when the patient inspired (fig 1). No other defects were observed. For further drainage a pleural tube was left in place, and a peritoneal tube was inserted to minimise the pressure on the diaphragm and the flow over the fistula. The next day the pleural tube leakage had nearly stopped and the chest radiograph showed only a small pleural effusion. Chemical pleurodesis was then performed with quinacrine (100 mg on three consecutive days) and the pleural drain was removed. Ten days after the removal of the peritoneal tube the patient was discharged in relatively good health without dyspnoea. After 2 years there are no clinical or radiological signs of recurrence of the pleural effusion.

DISCUSSION
Fontan operations for congenital heart abnormalities are frequently complicated by chronically raised systemic venous pressure. This can result in hepatomegaly, ascites and protein losing enteropathy. Pleural effusions in such patients can be...
the result of heart failure, hypoalbuminaemia, impaired lymphatic drainage, or leakage of ascites through the diaphragm. Unilateral left sided effusions are seldom seen. The venous pressure and albumin concentration in this patient on presentation were similar to those of the previous years but a large amount of pleural fluid was produced daily. We therefore hypothesised that the pleural effusion was caused by leaking of peritoneal fluid through the diaphragm. This has been described in patients with ascites due to liver cirrhosis, 2 in patients with peritoneal dialysis, 3 and those with ovarian hyperstimulation syndrome. 4 In these patients diaphragmatic defects or blebs have been found and it was postulated that peritoneal fluid could pass through them into the pleural cavity. Such passage can be shown with dyes or by nuclear imaging. In our patient thoracoscopy showed transdiaphragmatic leakage during inspiration caused by a ruptured diaphragmatic bleb forcefully squirting peritoneal fluid into the thorax. This shows how respiratory movements induce a transdiaphragmatic pressure gradient resulting in preferential flow of fluid from the peritoneum into the thoracic cavity. Such a mechanism has been hypothesised but has never been shown, and explains how only a small amount of peritoneal fluid results in a clinically significant pleural effusion. 1

Several procedures have been described to treat peritoneopleural fistulas. Clearly, the most efficacious treatment is ligation of the blebs (either by videothoracoscopy or thoracotomy) in combination with talc pleurodesis. This procedure was successful in 60% of 18 patients with a hepatic hydrothorax while talc insufflation alone was successful in 44% of patients. 5 The combination of videothoracoscopic surgery and talc pleurodesis was successful in all five patients with a peritoneopleural fistula due to peritoneal dialysis. 6

There are no studies reported of patients with right sided heart failure. As the cardiovascular status of our patient did not permit general or spinal anaesthesia, we used a combination of ascites drainage and chemical pleurodesis which resulted in satisfactory healing of the fistula and this persists to the present time.

REFERENCES