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CASE REPORT Open Access

Recurrent pleuroperitoneal leak caused by diaphragm blebs in a peritoneal dialysis patient: a case report with literature review

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Abstract

Background: Pleuroperitoneal leak is an uncommon but significant complication of peritoneal dialysis. Although the exact pathogenesis of pleuroperitoneal communication remains unclear, the pressure gradient between the thorax and abdominal cavity has been thought to play a major role.

Case presentation: A 48-year-old man with diabetes mellitus and hypertension who had been treated with continuous ambulatory peritoneal dialysis (1.5 L dwells four times a day) for 3 months was admitted because of massive right-sided pleural effusion. He underwent video-assisted thoracoscopic surgery for blebs on the diaphragm. Six weeks after diaphragmatic repair, he resumed peritoneal dialysis (1.5 L dwells four times a day) with once-a-week hemodialysis. Thereafter, pleural effusion was not significant on a chest radiogram. Six months after surgery, his dwell volume increased from 1500 to 2000 ml, and significant right-sided pleural effusion also developed.

Conclusion: Pleuroperitoneal leak caused by blebs can recur even after surgical treatment, and reducing the dwell volume may be effective for patients with pleuroperitoneal communication.

Keywords: Pleuroperitoneal communication, Video-assisted thoracoscopic surgery, Recurrent

Background

Pleural effusion frequently occurs in patients with end-stage renal disease. Common causes of pleural effusion in those patients include parapneumonia, uremic pleuritis, congestive heart failure, volume overload, tuberculosis, and malignancy [1]. Pleural effusion caused by pleuroperitoneal leaks is also a well-recognized complication of peritoneal dialysis (PD).

Although the prevalence of pleural effusion in PD patients varies, acute or subacute pleural effusion after starting PD is rare. The roles of diabetes mellitus on the pleural effusion after starting PD are not well documented. In Japan, the prevalence of acute pleural effusion was reported to be 1.6% [2]. The mechanism underlying the leaks is thought to be increased intra-abdominal pressure in the presence of an underlying congenital or acquired diaphragmatic defect [3]. Findings suggestive of pleuroperitoneal communication under PD include right-sided pleural effusion with elevated glucose concentration in the pleural fluid [4, 5]. In addition, a diaphragmatic defect is commonly found on peritoneal scintigraphy, although it may be difficult to diagnose such a small defect. Recently, the effectiveness of video-assisted thoracoscopic surgery (VATS) has been reported. However, the risks for the recurrence of pleuroperitoneal leak after VATS have not been documented.

We herein report a PD patient with a slight but significant peritoneal-pleural leak that was difficult to diagnose and recurrent after VATS.

Case report

A 48-year-old man with end-stage renal failure was admitted because of massive right-sided pleural effusion. He had received anti-diabetic and anti-hypertensive agents for more than 7 years. Four months before this admission, he developed end-stage renal failure and started hemodialysis

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therapy. One month later, his renal replacement therapy was switched from hemodialysis to PD. He was treated with standard continuous ambulatory PD (CAPD) with overnight icodextrin dwell (using 1.5 L of dialysis fluid containing 1.5% glucose three times and 1.5 L of dialysis fluid containing 7.5% icodextrin). Three months after the initiation of CAPD, he felt dyspnea, and massive right-sided pleural effusion was found on chest radiography (Fig. 1a). Abdominal ultrasonography found no findings that suggested liver cirrhosis. The pleural fluid-to-serum protein ratio was 0.03 (total protein concentrations of pleural fluid and serum: 0.2 and 6.5 g/dL, respectively), and the pleural fluid glucose level was 249 mg/dl. Renal replacement therapy was switched from CAPD to hemodialysis. Thereafter, the pleural effusion disappeared. These findings indicated that the massive pleural effusion in this patient was caused by pleuroperitoneal communication. He was therefore referred to our hospital for a further examination.

On admission, a physical examination revealed a blood pressure of 123/55 mmHg and no edema in his lower legs. His urine output was about 500 ml/day. Laboratory examinations showed the following: total protein 6.8 g/dL, serum albumin 3.7 g/dL, total cholesterol 226 mg/dl, triglycerides 133 mg/dl, serum creatinine 8.0 mg/dL, blood urea nitrogen 39 mg/dL, aspartate aminotransferase 47 U/L, alanine aminotransferase 26 U/L, and C-reactive protein 0.03 mg/dL. Hematological tests showed hemoglobin 13.3 g/dL, white blood cell count 4.800/μL, and platelet count 164,000/μL. His thyroid function was normal. His BNP level was elevated to 534 pg/ml, although his cardiac contraction was preserved (ejection fraction, 64% on cardiac ultrasound), and no valvar diseases were found. Pleural effusion was not significant on a chest radiogram.

To determine the cause of the pleural effusion during CAPD, he was treated with combination therapy with standard hemodialysis (thrice weekly) with overnight dwell of 2 L of dialysis fluid containing 1.5% glucose. Four days after starting the combination therapy, right-sided pleural effusion was found on a chest radiogram without body weight gain and pretibial edema. To confirm the presence of peritoneal-pleural leak by scintigraphy, a mixture of 370 MBq Tc-99 m MAA in 2 L of dialysis fluid was infused into the peritoneal cavity through a Teckhoff catheter. However, no abnormal tracer accumulation was detected in the thoracic cavity. Although peritoneal-pleural leak was not confirmed on peritoneal scintigraphy, we attempted to perform repair and/or reinforcement of his right diaphragm by VATS in accordance with the patient's strong desire to continue PD. During surgery, blebs were found on the central tendon of the diaphragm by pneumoperitoneum (Fig. 2). We ligated the origin of the bleb and tightened the bleb with a vertical mattress suture. We then affixed fibrin sealant patches and polyglycolic acid sheets to the suture. Finally, we laid down exogenous fibrin glue. After the reinforcement, the blebs were no longer detected even when pneumoperitoneum was performed.

Six weeks after surgery, he resumed PD (using 1.5 L of dialysis fluid containing 1.5% or 2.5% glucose three times and 1.5 L of dialysis fluid containing 7.5% icodextrin). He also received hemodialysis once a week because the fluid removal by PD was insufficient, and right-sided pleural effusion was not significant (Fig. 1b). However, 6 months after surgery, significant right-sided pleural effusion recurred in association with the changes in his dwell volume for PD from 1500 to 2000 ml (Fig. 1c). His dwell volume was then increased from 2000 to 1500 ml, thereby reducing his plural effusion, and he was treated with combined PD (using 1.5 L of dialysis fluid containing glucose three times and 1.5 L of dialysis fluid containing 7.5% icodextrin) and hemodialysis (once a week).

Discussion

In Japan, the number of patients who underwent PD was 9021 at the end of 2016, which was 2.7% of the entire dialysis patient population. The number of new dialysis patients was 39,344, and 1946 of those (4.9%) started renal replacement therapy using PD. In patients with end-stage renal failure who start PD, massive refractory pleural effusion caused by peritoneopericardial communication is rare but a

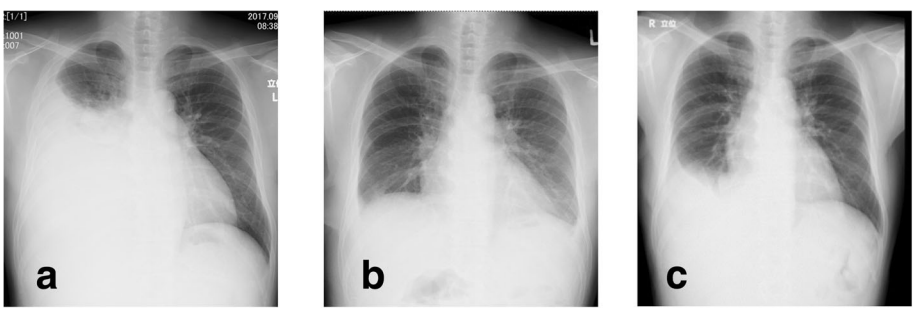

Fig. 1 Changes in the pleural effusion. **a** At the first admission after starting continuous peritoneal dialysis. **b** After VATS. He was treated with continuous peritoneal dialysis using 1.5 L of dialysis fluid and hemodialysis (once a week). **c** After VATS. He was treated with continuous peritoneal dialysis using 2 L of dialysis fluid and hemodialysis (once a week)

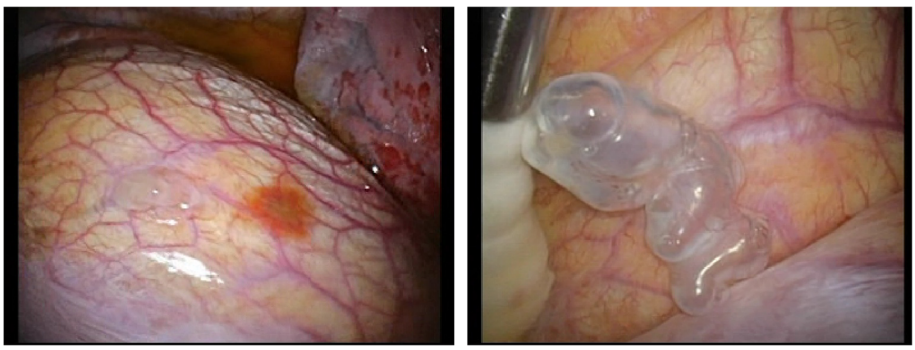

Fig. 2 Surgical findings. Bleb on the pneumoperitoneum before (left) and after (right) pneumoperitoneum. Bleb located on the central tendon of the diaphragm

major cause of discontinuing PD. In the present case, the pleural fluid-to-serum protein ratio was 0.03 and serum albumin level was within normal range (3.7 g/dL) with normal ultrasound image of the liver. His cardiac contraction was preserved, and the right-sided pleural effusion was found after starting the combination therapy with hemodialysis and peritoneal dialysis without body weight gain and pretibial edema. These findings indicated that other causes of transudative pleural effusion than the peritoneopericardial communication, such as liver cirrhosis, hypoalbuminemia, congestive cardiac failure, or other causes of fluid overload were not associated with the pleural effusion.

It has been well recognized that pleuroperitoneal defects result in pleural effusion in PD patients. In PD, a dialysate is infused through a tube into the peritoneal cavity. Thus, the intra-abdominal pressure is elevated, and accordingly, a pressure gradient is generated between the intra-abdomen and intra-thorax [6]. There are a number of reasons a leak could occur, including leakage around the major vessels and/or esophagus, diaphragmatic foramina, the diaphragmatic lymphatics, and the thoracic duct [7, 8]. Blebs in the diaphragm, which are very thin and lacking in tissues such as the tendons and skeletal muscles, also transfer the dialysate from the abdomen to the chest as a one-way valve [9].

Pleural effusion caused by pleuroperitoneal communication is mainly on the right side, accounting for 88% of cases, with 8% of cases left-sided and 4% bilateral [2]. There are several tests to diagnose pleuroperitoneal communication. Diagnostic thoracentesis and pleural fluid analyses are usually performed. If the pleural fluid glucose level is greater than 300 mg/dL, it indicates that the pleural fluid is accumulated due to pleuroperitoneal communication [5]. However, some patients show a relatively low glucose gradient, as glucose is absorbed by the pleural mesothelium [7]. We should therefore doubt pleuroperitoneal communication if the pleural fluid glucose level is higher than that in the serum [4]. In our case, the glucose level in the pleural fluid was 249 mg/dl, and the blood glucose levels ranged from 100 to 250 mg/dl. In addition, positive tests of scintigraphy or computed tomography (CT) peritoneography for the pleuroperitoneal communication are obtained in only half or a third of cases, respectively [6, 7]. Therefore, even if the pleural fluid glucose level is < 300 mg/dl and/or imaging test do not prove peritoneal-pleural leak, pleuroperitoneal communication may still be present in PD patients with right-sided pleural effusion.

The exact mechanism underlying spontaneous pleuroperitoneal communication remains unknown. In our case, blebs were found on the diaphragm. In cases of spontaneous pneumothorax, spontaneous rupture of a subpleural bleb or of a bulla may be the major causes [8]. The recurrence rate of spontaneous pneumothorax after VATS was reported to be 11.4%. The mechanisms underlying recurrence were reported as follows: only a minority of blebs are actually ruptured at the time of surgery, although the lesions are present [9]. In addition to the influence of the blebs remaining after surgery, increased intra-abdominal pressure may play a major role in the genesis of pleural leak [3]. Indeed, the pleural fluid volume changed in association with the dwell volume of dialysate.

Conservative treatment of hydrothorax related to pleuroperitoneal communication in PD patients has shown limited success [2, 10]. Chou et al. reviewed 10 population-based case series in which 44 of the 104 cases (42%) were unable to resume PD [11]. Therefore, the establishment of treatments for pleuroperitoneal communication in patients with end-stage renal failure has been required. The efficacy of chemical pleurodesis under video-assisted thoracoscopic guidance for hydrothorax has been confirmed, and the success rate was reported to be 90% [10]. The effectiveness of repair and/or reinforcement therapy performed by VATS has also been reported [12, 13]. However, the risk for the recurrence of pleuroperitoneal leak after VATS is unclear. In the present case, the volume of right-sided pleural effusion changed in accordance with the dwell volume of dialysate. To prevent the recurrence of pleuroperitoneal leak, we should pay particular attention to

the dwell volume, especially in cases of pleuroperitoneal leak caused by blebs.

Mini review: procedures used to demonstrate a pleuroperitoneal leak

Glucose concentration in the pleural fluid

This is a low-cost procedure. However, it is difficult to get a definitive diagnosis by glucose concentrations in the pleural fluid (and serum). The pleural fluid-to-serum glucose gradients were reported to be variable from 2 to 1885 mg/dl in PD patients with diagnosed pleuroperitoneal communication [14]. Although plural to serum glucose ratio > 1 was reported to be useful, serum glucose levels were also variable in patients with diabetes.

Imaging tests (CT peritoneography or scintigraphy)

In a case study, CT peritoneography showed a 33% sensitivity in identifying pleuroperitoneal leak and sensitivity of radionuclide scans (Tc-99 m DTPA) was reported to be 40–50% [7, 15]. These results indicate that these imaging tests were inadequately appraised. In addition, there are some disadvantages in these procedures. Contrast CT peritoneography may add the risk of nephrotoxicity and reduce the residual renal function in PD patients. Further, in Japan, medical costs of scintigraphy for a pleuroperitoneal leak are not covered by health insurance.

VATS

This invasive procedure can detect diaphragmatic defects or blebs under the direct vision. However, the usefulness of VATS has been well documented in the treatment, but not in the diagnosis for a pleuroperitoneal leak in PD patients.

These three procedures differ in risk, cost, availability, and reliability. As in the present case, VATS with pneumoperitoneum may be useful in a difficult disease to diagnose.

Conclusion

Pleuroperitoneal leak caused by blebs can recur even after surgical treatment, and reducing the dwell volume may be effective for patients with pleuroperitoneal communication.

Abbreviations

CAPD: Continuous ambulatory peritoneal dialysis; CT: Computed tomography; PD: Peritoneal dialysis; VATS: Video-assisted thoracoscopic surgery

Availability of data and materials

Data will be made available by the corresponding author upon request.

Authors' contributions

YH and TU are responsible for the manuscript. YH NK, KI, HK, HM, and TU provided the best medical care for the patient and contributed to the collection of data of the present case. All authors read and approved the final manuscript.

Ethics approval and consent to participate

According to the Ethical Guidelines for Medical and Health Research involving Human Subjects in Japan, ethics approval is not necessary for case reports.

Consent for publication

Written informed consent was obtained from the patient for the publication of this case report.

Competing interests

The authors declare that they have no competing interests.

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