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Case Report

Effective Management of Peritoneal Dialysis-Associated Hydrothorax in a Child: A Case Report

Cahyani Gita Ambarsari[a] Evita Karianni Bermanshah[a]
Muhammad Arza Putra[b] Farhan Haidar Fazlur Rahman[a]
Sudung Oloan Pardede[a]

[a]Department of Child Health, Faculty of Medicine, Universitas Indonesia, Cipto Mangunkusumo Hospital, Jakarta Pusat, Indonesia; [b]Department of Surgery, Faculty of Medicine, Universitas Indonesia, Cipto Mangunkusumo Hospital, Jakarta Pusat, Indonesia

Keywords
End-stage renal disease · Peritoneal dialysis · Pleural effusion · Pleurodesis

Abstract
Peritoneal dialysis (PD) confers many advantages, including a better quality of life for children with end-stage renal disease; however, the procedure is associated with several complications, including pleuroperitoneal leaks. Here, we report an unusual case of hydrothorax caused by long-term PD in a child, which was further complicated by pneumonia. A 9-year-old boy who had received CAPD for 22 months presented with dyspnea, swelling, and increased body weight. Chest tube drainage yielded 500 mL of transudative fluid. Computed tomography peritoneography revealed increased outflow from the peritoneum to the pleural cavity. PD was suspended, and hemodialysis (HD) was initiated. Video-assisted thoracoscopic surgery was performed; however, because the patient had pneumonia during hospitalization, pleural adhesions with a septated appearance occurred. This resulted in difficulties identifying pleuroperitoneal fistula (PPF). Right pleural effusion resolved following pleurodesis using bleomycin.

KARGER

Cahyani Gita Ambarsari
Department of Child Health, Faculty of Medicine, Universitas Indonesia
Cipto Mangunkusumo Hospital, Diponegoro 71
Jakarta Pusat 10430 (Indonesia)
cahyani.ambarsari@ui.ac.id, cahyani.peds@yahoo.com

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Regular HD was performed for 10 weeks, and PD was subsequently reinitiated. There was no recurrence of hydrothorax during long-term follow-up. We suspect that the underlying mechanism of hydrothorax in our patient was associated with a PPF that formed either due to a congenital diaphragmatic defect or an acquired defect, resulting in dialysate leakage. Our case demonstrates that a temporary switch from PD to HD, accompanied by pleurodesis, may help resolve hydrothorax that occurs as a complication of long-term PD.

Introduction

Hydrothorax secondary to a pleuroperitoneal fistula (PPF) is a rare complication of long-term peritoneal dialysis (PD); its incidence rate is 0.53% in children. While the underlying mechanism remains unknown, it is speculated to be associated with a pleuroperitoneal communication [1, 2].

We present a pediatric case of hydrothorax that occurred as a complication of long-term PD; hydrothorax was confirmed by increased outflow to the thoracic cavity observed on computed tomography (CT) peritoneography with contrast. A PPF was not detected during video-assisted thoracoscopy surgery (VATS) because the case was complicated by pneumonia, with pleural adhesions showing a septated appearance.

Case Presentation

A 9-year-old Indonesian boy on CAPD was admitted to the emergency unit of our center due to dyspnea, swelling, and increased body weight. There was no fever, cough, or chest pain. Two years previously, he was diagnosed with end-stage renal disease (ESRD) secondary to bilateral renal hypoplasia; since then, he had been receiving CAPD comprising five cycles of 1-L exchanges with 1.5% dextrose-based solution for 22 months. Furthermore, he had no medical history of heart disease, peritonitis, hernia, thoracic and abdominal trauma, pneumonia, or thoracic and abdominal surgery, except for Tenckhoff catheter insertion. During the previous 3 days, his body weight had increased by 4 kg (12%), and he experienced ultrafiltration failure because although PD fluid at 1 L per cycle was being infused as usual, the effluent drainage was only approximately 0.6 L per cycle.

Physical examination revealed a respiratory rate of 34 breaths/min, heart rate of 120 beats/min, and blood pressure of 147/86 mm Hg. Oxygen saturation was 92% with nasal oxygen at 2 L/min. Auscultation of the lungs revealed diminished vesicular sounds in the right chest wall. Cardiac findings were normal. Arterial blood gas analysis results were as follows: pH: 7.467, pCO_2: 4.51 kPa, pO_2: 22.52 kPa, bicarbonate: 24.8 mmol/L, and base deficit: 2 mmol/L. His hemoglobin level was 131 g/L, albumin level was 34.6 g/L, BUN level was 7.08 mmol/L, and creatinine level was 840.33 μmol/L. Chest radiograph revealed right pleural effusion and bilateral pulmonary infiltrates (Fig. 1a). The chest tube drained 500 mL of transudative fluid. A comparison of serum, pleural fluid, and peritoneal fluid parameters has been presented in Table 1.

After the excessive pleural fluid was completely removed, PD was continued via an installed chest tube (Fig. 1b). Intravenous cefotaxime was administered to treat pneumonia that occurred as an additional complication. Recurrent pleural effusion was subsequently found along with ultrafiltration failure. Pleural fluid culture did not show any microbial growth. Hydrothorax secondary to PPF was suspected, and PD was suspended from the 8th day of hospitalization. Hemodialysis (HD) was subsequently initiated.

CT peritoneography was performed on the 9th day using 300 mg/mL of iohexol contrast agent immersed in PD fluid. Serial examinations at 30 and 180 min after administering the contrast agent showed excessive fluid in the right pleural cavity (Fig. 1c–h). PPF in the right posterolateral side was suspected.

The patient underwent VATS on the 20th day of hospitalization; however, no fistula was detected in the right chest cavity. Instead, adhesions between the parietal and visceral pleura with septated appearance were observed (Fig. 2a, b). Pleurodesis was performed using 20 mL of bleomycin for the right diaphragm. Following this, pleural effusion of the right lung resolved (Fig. 3a), and the patient underwent regular HD for 10 weeks. Subsequently, PD was gradually initiated until achieving 1 L of 1.5% dextrose-based dialysate solution with five daily exchanges. A 3-year follow-up showed normal PD without any recurrence of hydrothorax (Fig. 3b).

Discussion/Conclusion

This paper describes a pediatric case of hydrothorax that occurred as a complication of long-term PD. PD is the recommended treatment modality of renal replacement therapy for children with ESRD. It confers a better quality of life [3]; however, it is associated with noninfectious complications, such as catheter obstruction or malfunction, leakage, abdominal wall defect, PPF, hydrocele, ultrafiltration failure, or fluid overload [4].

Symptoms of hydrothorax usually involve respiratory distress, particularly dyspnea [1, 2] – which was present in our patient – as well as diminished vesicular sounds on auscultation and pleural effusion on chest radiograph. However, the underlying mechanism of hydrothorax secondary to PD remains unknown. Hydrothorax is postulated to be associated with a PPF that is formed secondary to a congenital diaphragmatic defect or an acquired defect, resulting in the movement of dialysate from the peritoneum to the pleural cavity [1, 2]. Alteration in the pleuroperitoneal pressure gradient secondary to PD that occurred over a 21-month period in our patient may have triggered PPF [5].

CT peritoneography is advantageous for depicting the entire peritoneal and thoracic cavity with minimal costs compared with magnetic resonance peritoneography [6]. Peritoneal scintigraphy may be performed using technetium-99m macroaggregated albumin (^{99m}Tc-MAA) as the contrast agent [5]. We used iohexol as the contrast agent, which highlights a characteristic imaging feature of PPF. Possible defects in the pleura and diaphragm were identified using VATS, but no fistula was observed in the right chest cavity. Instead, adhesions with a septated appearance, presumably secondary to pneumonia, made it difficult to identify the fistula.

Although talc offers better results – as it can inhibit angiogenesis and prevent recurrent pleural effusion [7] – bleomycin was used for pleurodesis in our patient because of the

unavailability of talc. Furthermore, bleomycin is less toxic compared with other agents such as povidone iodine [8]. Although bleomycin had a higher cost compared with talc (USD 796 vs. USD 488), bleomycin had a success rate of 87.6% in preventing the recurrence of effusion in comparison with talc, which was 90.2% [9]. There have been 5 cases of pleurodesis during 2016–2019 at our center in which bleomycin was used in adults with catamenial pneumothorax (n = 1) and tumor metastasis-associated spontaneous pneumothorax (n = 4) as primary diseases. No side effects or recurrence of pneumothorax was observed during the 1- to 3-year follow-ups. This is the first case of pleurodesis using bleomycin as well as the first case of pleurodesis for suspected PPF in a pediatric patient under long-term PD at our center.

Our findings suggest that discontinuing PD can prevent the occurrence of an expanding effusion. Temporary HD for 4–16 weeks has been previously recommended [10]. Accordingly, we performed HD for 10 weeks and subsequently reinitiated PD. Long-term follow-up revealed no recurrence of hydrothorax. In conclusion, we showed that diagnosing PPF via CT peritoneography using iohexol contrast, followed by a temporary switch of PD to HD, and performing pleurodesis using bleomycin is a potentially valid method for treating a patient with hydrothorax that occurs as a complication of long-term PD in limited settings.

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Statement of Ethics

The research was ethically conducted in accordance with the World Medical Association Declaration of Helsinki. Written informed consent was obtained from the caregiver for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor of this journal.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Author Contributions

C.G.A. and F.H.F.R. performed the literature search, data collection, data analysis, and data interpretation and wrote the first draft of the manuscript. E.K.B. and M.A.P. performed the data collection and data analysis, reviewed the manuscript, and approved the final version. S.O.P critically reviewed the manuscript. All authors read and approved the final manuscript.

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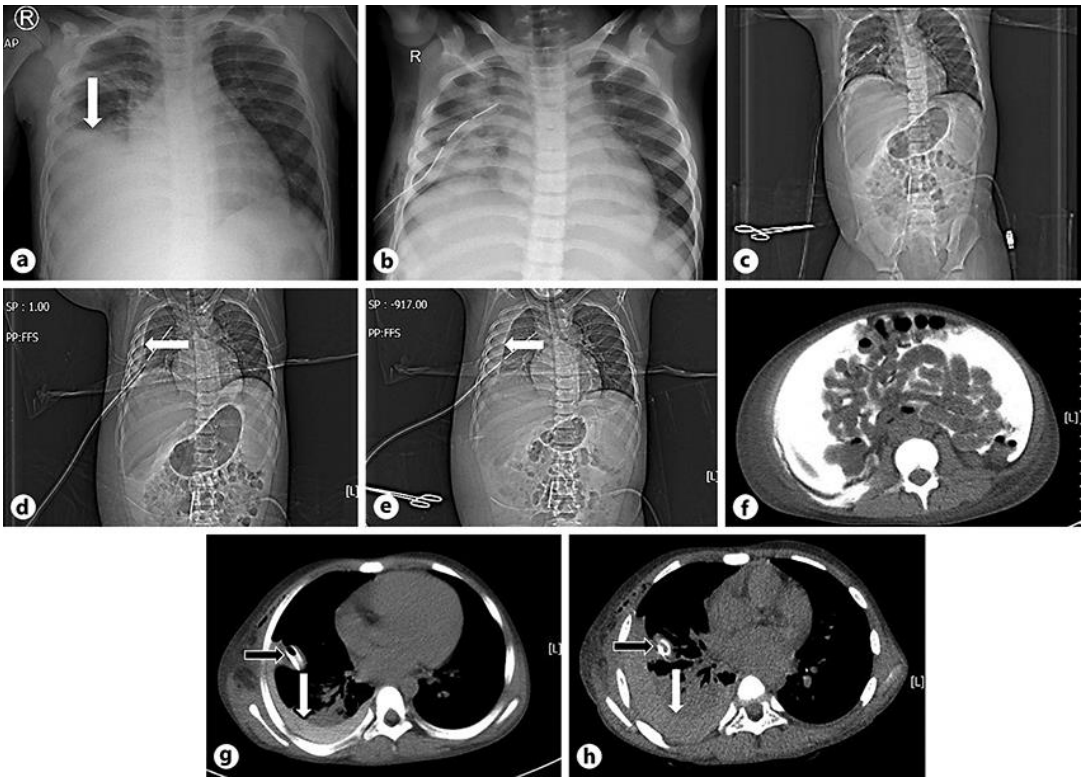

Fig. 1. Imaging series of a 9-year-old male patient who experienced hydrothorax while on CAPD. **a** An initial chest radiograph showed right pleural effusion. **b** The pleural effusion resolved, and PD was performed with chest tube still attached. Note the presence of pneumonia. **c** Chest radiograph immediately prior to CT peritoneography. Chest radiograph shows increased presence of fluid in the right pleural cavity at the 30th min (**d**) and 180th min (**e**) (white arrows). **f** CT peritoneography of the peritoneum surrounded by peritoneal dialysis solution containing contrast media shows increased presence of fluid in the right pleural cavity at the 30th min (**g**) and 180th min (**h**) (white arrows), with black arrows indicating the position of the chest tube.

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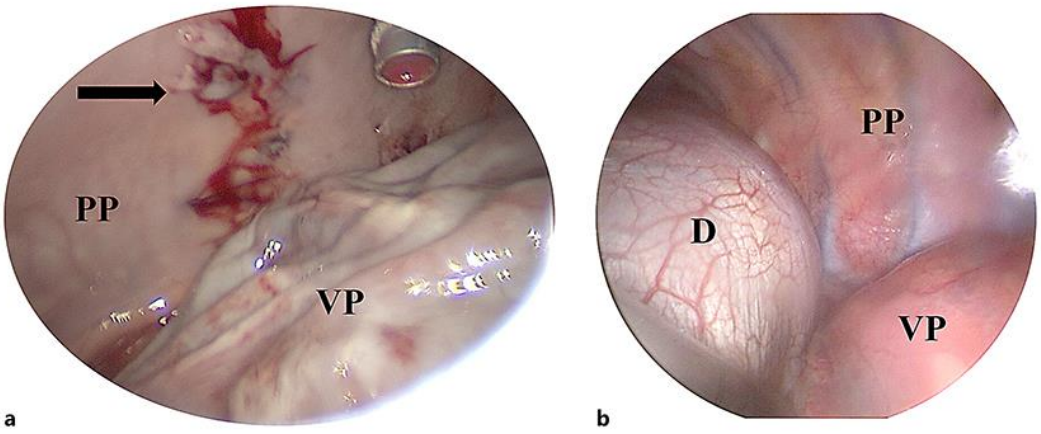

Fig. 2. **a** VATS shows inflammation in the region following septa removal prior to adhesiolysis. **b** VATS showed no defects over the diaphragm, and fistula was not seen.

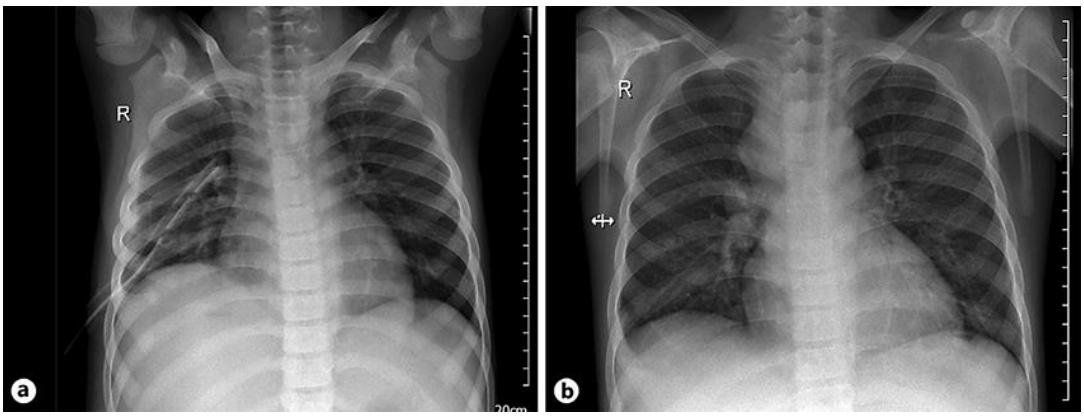

Fig. 3. **a** First follow-up chest radiograph after PD suspension and HD initiation showed reduction of pleural effusion. **b** The 3-year follow-up chest radiograph showed complete resolution of pleural effusion with no recurrence.

Table 1. Serum, peritoneal, and pleural fluid biochemistry

Specimen	Glucose, mmol/L	LDH, μkat/L	Protein, g/L	Cell count, /μL
Reference	3.9–6.1	1.7–3.4	62–85	
Serum	5.49	12.11	54	
Peritoneal fluid	38.57	0.1	2	2
Pleural fluid	8.27	1.02	3	26

LDH, lactate dehydrogenase.