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BRIEF REPORT

Peritoneal–pericardial communication in an adolescent on peritoneal dialysis

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Abstract
Background Dialysate leakage into the pericardium is a rare but potentially life-threatening complication of peritoneal dialysis (PD). There has been one reported pediatric case of spontaneous peritoneo-pericardial fistula in a 2-year-old boy with tissue fragility due to malnutrition and two reported adult cases in PD patients with a history of previous cardiac surgery and/or pericardiocentesis.
Case-Diagnosis/Treatment We describe a 15-year-old girl with end-stage renal disease secondary to granulomatosis with polyangiitis, with recurrent pericardial effusions secondary to a peritoneo-pericardial fistula while on continuous cycling peritoneal dialysis (CCPD). She had previously presented with chylous pericardial effusion that required pericardiocentesis and subsequently developed recurrent pericardial effusions when she was commenced on CCPD 9 months later. Pericardial fluid chemistry revealed a sterile, serous fluid containing 15.1 mmol/L of glucose and <0.11 mmol/L of triglycerides. Peritoneal scintigraphy with Tc-99m labeled sulfur colloid injected intra-peritoneally confirmed the presence of a peritoneo-pericardial fistula. The pericardial effusions resolved upon switching the patient to hemodialysis (HD).
Conclusions Our case of recurrent pericardial effusions in a child on PD secondary to a peritoneo-pericardial fistula highlights the need for close follow-up in patients with a history of previous pericardiocentesis who are commenced on PD.

Keywords Dialysate leakage · Pericardial effusions · Peritoneal fluid scintigraphy · Vasculitis

Case summary

We describe a 15-year-old girl with granulomatosis with polyangiitis (GPA) who presented with pulmonary hemorrhage, active urinary sediment and normal creatinine. Renal biopsy demonstrated a moderately severe pauci-immune, crescentic glomerulonephritis with focal segmental fibrinoid necrosis. She was anti-neutrophil cytoplasmic antibody positive (1:320) with an elevated proteinase 3 (1:80) and normal myeloperoxidase titre (1:2). Her pulmonary disease responded to combined therapy of intravenous (IV) pulse methylprednisolone and cyclophosphamide.

Following 2 months of maintenance immunosuppressive therapy, she developed a large pericardial effusion. Subxiphoid pericardiocentesis drained 450 ml of chyle, and a pericardial drain remained in place for 4 days, with resolution following treatment with IV octreotide. Ongoing echocardiographic follow-up over 6 months confirmed the absence of a pericardial fluid re-accumulation.

Despite therapy, her renal disease progressed to chronic kidney disease stage 5 over the next 6 months, with repeat renal biopsy demonstrating widespread glomerulosclerosis. She commenced peritoneal dialysis (PD) 11 months after

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initial presentation. One week following the initiation of nighttime intermittent PD (NIPD) (900 ml/m^2 fill volumes—Dianeal® 1.5 %), a routine chest X-ray revealed an enlarged cardiac silhouette, and echocardiogram confirmed a large pericardial effusion. She underwent a second sub-xiphoid pericardiocentesis/insertion of a drain. The 500 ml of aspirated pericardial fluid appeared to be serous (fluid protein <20 g/L, fluid glucose 6.3 mmol/L, serum glucose 6.8 mmol/L) and had a nucleated cell count of 4400×10^6 /L, but was sterile and negative for adenovirus, enterovirus and echovirus. Her C-reactive protein and erythrocyte sedimentation rate were both moderately elevated, and she had mild lymphopenia and anemia. Her anti-neutrophil cytoplasmic antibody titers were negative. The differential diagnoses at the time included an inflammatory pericarditis secondary to her GPA (raised inflammatory markers and pericardial fluid with inflammatory cells), pericarditis secondary to infection and uremic pericarditis, although the latter was deemed unlikely owing to her considerable residual renal function and a sufficient PD prescription. As pericarditis secondary to infection also seemed unlikely based on repeated negative viral and bacterial cultures she was treated with steroids (oral prednisone 1 mg/kg daily).

She continued on PD for the next 48 h, with significant volumes of glucose-containing solution draining from the pericardial drain (fluid protein <20 g/L, fluid glucose 16.8 mmol/L, serum glucose 4.6). As there were concerns that peritoneal fluid was tracking up into the pericardium, a 12.5-French non-cuffed hemodialysis (HD) neck catheter was inserted, her PD was held (abdomen drained) and she commenced HD four times weekly, achieving urea reduction ratios of 70–75 % on each occasion. Serial echocardiography following removal of the drain documented no re-accumulation over the next 2 weeks while on HD.

NIPD was resumed with a fill volume of 900 mL/m^2, 8 cycles over 10 h with Dianeal® 1.5 %. The fill volume was increased gradually over the next 5 days to 1100 mL/m^2. Routine surveillance echocardiography 1 week after restarting NIPD demonstrated a significant pericardial effusion, requiring further pericardial drainage (fluid volume 600 mL, fluid protein <20 g/L, fluid triglycerides <0.11 mmol/L, fluid glucose 15.1 mmol/L, serum glucose 4.4 mmol/L).

Peritoneal scintigraphy was planned in order to demonstrate the presumed peritoneal–pericardial connection. Briefly, 222 MBq of Tc-99m sulphur colloid was injected into 1500 mL of peritoneal dialysate and infused into the patient's peritoneum via the Tenckhoff catheter. Dynamic images were taken up to 60 min followed by static images. A subsequent dynamic acquisition sequence and single-photon emission computed tomography (SPECT), as well as 2.5-h delayed images were also obtained. Initially, activity was only seen in the peritoneum. However, during the dynamic acquisition, activity was visualized in the region of the pericardium (Fig. 1) and subsequently confirmed on the second dynamic acquisition series and SPECT.

Following the positive test she was switched back to HD, and the pericardial effusion resolved echocardiographically over 1 month, without any need for further drainage. A repeat nuclear study 2 weeks following the switch to HD, prior to the removal of her PD catheter, confirmed a persistent peritoneal–pericardial communication. She has remained on HD four times weekly with no further re-accumulation of pericardial fluid.

Discussion

Leaks into the pleural cavity via congenital or traumatic diaphragmatic defects are potentially life-threatening and have a reported incidence of 1.6 % in adults on PD [1]. However, dialysate leakage into the pericardium itself is rare and has, to the best of our knowledge, only been described previously in two adult PD patients with a history of prior cardiac surgery and pericardiocentesis and in a 2-year-old child on PD who developed a spontaneous peritoneo-pericardial fistula secondary to obvious tissue fragility due to severe malnutrition [2–4]. In a group of 1506 children followed over 10 years, Dufek et al. recently reported a prevalence of 0.13 % for pericardial effusions secondary to peritoneo-pericardial fistulae and 0.6 % for pleural effusions secondary to pleuro-peritoneal fistulae [5]. When pericardial effusions do occur, they tend to become clinically apparent early upon PD initiation [1]. Risk factors include excessive and/or rapid increase in fill volume, previous peritonitis, cardiac and/or abdominal surgery and malnutrition [2, 5].

Our patient initially developed a chylous pericardial effusion, presumably secondary to her underlying vasculitis. This led to the need for pericardiocentesis and the placement of a pericardial drain [6]. While cardiac involvement in GPA is uncommon, affecting 6–9 % of children and adults [7, 8], pericarditis is the most commonly recognized form of such involvement [9].

Patients with pericardial effusions often present with minor symptoms, such as cough, but may also complain of abdominal and chest pain, dyspnoea and/or life-threatening cardiac tamponade depending upon the hemodynamic significance of the fluid collection [2]. Our patient was asymptomatic despite having moderate to large pericardial effusions and was diagnosed solely on the basis of an enlarged cardiac silhouette on a routine chest X-ray.

All subsequent occurrences of our patient's pericardial effusion developed within 1 week of PD initiation, with the first manifestation occurring upon PD initiation 9 months post-pericardiocentesis. The non-chylous, sterile fluid, associated with relatively elevated levels of glucose in comparison to her serum glucose, raised valid concerns about potential tracking

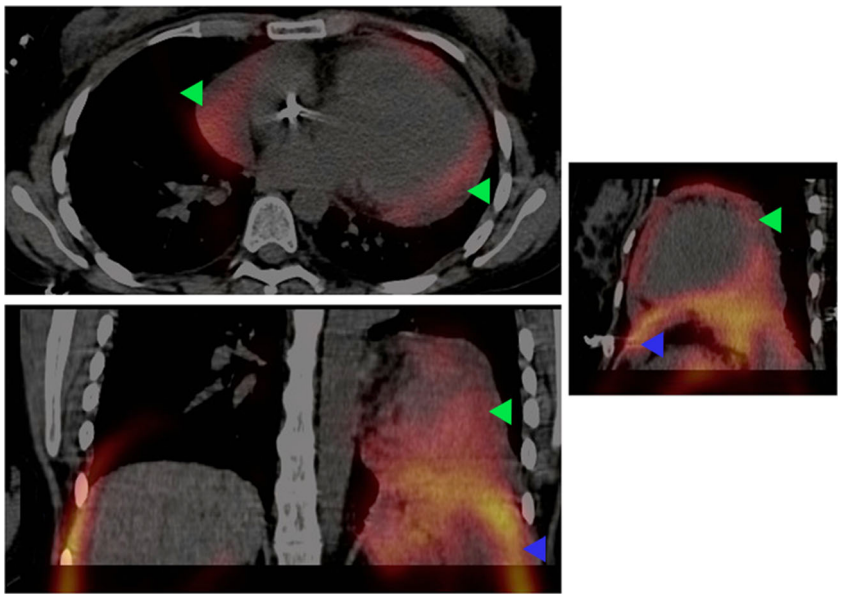

Fig. 1 Colloid (222 MBq) injected into 1500 ml of peritoneal dialysate (*blue arrowheads*) revealed the presence of activity in the pericardium (*green arrowheads*)

of peritoneal dialysate from the peritoneum along the tract created by the pericardial drain into the pericardial sac.

As such, our patient was initially switched to HD during the first recurrence of the effusion following the first pericardial drain placement post-PD. Of note, during this intensive time on HD, weekly echocardiography did not show any significant re-accumulation of pericardial fluid.

Upon the resumption of PD the immediate presence of pericardial effusion, the analysis of which demonstrated supra-physiologic levels of glucose (14.8 mmol/L), was in keeping with a peritoneal–pericardial communication that was confirmed with peritoneal scintigraphy (Fig. 1). We postulate that our patient developed this persistent tract following her initial pericardiocentesis and drain placement (9 months prior) in the context of an inflammatory disease, steroid use and delayed wound healing [3, 4]. Her PD exchange volume was not excessive and she was not malnourished.

While we also entertained the possibility of a presumed congenital pericardial defect resulting in the communication between the pericardial sac and mediastinum, we have been unable to find any mention of any such congenital anatomical defect(s) in the medical literature or textbooks. Also, the absence of fluid collection outside of the pericardium makes such a defect unlikely. In addition, our patient did not have a pleural effusion, which is often associated with peritoneal dialysate leaking through diaphragmatic defects [1].

Many techniques including intraperitoneal instillation of methylene blue, computerized tomography (CT) peritoneography [10], magnetic resonance (MR) peritoneography [2, 11] and peritoneal scintigraphy [12] can be used to diagnose problems related to dialysate fluid leakage. Methylene blue is an irritant and is associated with chemical peritonitis [13]. Moreover, it would require the placement of a pericardial and/or pleural drain to demonstrate an abnormal communication. CT peritoneography is capable of delineating the peritoneal cavity clearly but would require the use of intraperitoneal contrast; and the patient, especially the pediatric patient, is also exposed to a considerable radiation dose [12]. While MR peritoneography can also provide a detailed anatomical distribution of dialysate without the need for intraperitoneal contrast and ionizing radiation, its higher cost is a potential drawback and one may not be able to track fluid from one to another discrete compartment. MR imaging facilities may also not be as widely available in many centers.

The usefulness of peritoneal scintigraphy in diagnosing peritoneal leaks has been illustrated in various case reports [12, 14]. The radiopharmaceutical agent used in peritoneal scintigraphy has to consist of large, non-absorbable particles to prevent it from being transferred across the peritoneal membrane and communicating with lymphatics. Several agents, including Tc-99m sulfur colloid, Tc-99m MAA and Tc-99m-DTPA, have been used. This technique is associated with a sensitivity ranging between 40 and 50 % [15]. To the best of our knowledge, our case is the first to highlight its use in diagnosing a peritoneal–pericardial fistula in a pediatric patient.

In summary, this case highlights pericardial effusions as a rare but potentially life-threatening complication of PD in a patient with potential peritoneo-pericardial communication, as might be seen in a post-operative heart patient with acute kidney injury or in a patient who has undergone pericardiocentesis. It can occur late after the cardiac procedure (9 months post-pericardiocentesis and drain in our case) and tends to occur within a few weeks of PD initiation. We illustrate the utility of peritoneal fluid scintigraphy in diagnosing an abnormal peritoneo-pericardial fistula in a pediatric patient. Based on our experience, we suggest conversion to HD for a prolonged period of time to likely be the best therapeutic option.

Conflict of interest The authors declare that they have no competing interests.

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Patient consent Obtained.

References

1. Nomoto Y, Suga T, Nakajima K, Sakai H, Osawa G, Ota K, Kawaguchi Y, Sakai T, Sakai S, Shibata M (1989) Acute hydrothorax in continuous ambulatory peritoneal dialysis—a collaborative study of 161 centers. Am J Nephrol 9:363–367
2. Borzych D, Ley S, Schaefer F, Billing H, Ley-Zaporozhan J, Schenk J, Schmidt CP (2008) Dialysate leakage into pericardium in an infant on long-term peritoneal dialysis. Pediatr Nephrol 23: 335–338
3. Nather S, Anger H, Koall W, Reis A, Mukhtar B, Grimm C, Drescher J, Osten B (1996) Peritoneal leak and chronic pericardial effusion in a CAPD patient. Nephrol Dial Transplant 11:1155–1158
4. Hou CH, Tsai TJ, Hsu KL (1994) Peritoneopericardial communication after pericardiocentesis in a patient on continuous ambulatory peritoneal dialysis with dialysis pericarditis. Nephron 68:125–127
5. Dufek S, Holtta T, Fischbach M, Ariceta G, Jankauskiene A, Cerkauskiene R, Schmitt CP, Schaefer B, Aufricht C, Wright E, Stefanidis CJ, Ekim M, Bakkaloglu S, Klaus G, Zurowska A, Vondrak K, Vande Walle J, Edefonti A, Shroff R, European Paediatric Dialysis Working Group (2015) Pleuro-peritoneal or pericardio-peritoneal leak in children on chronic peritoneal dialysis—a survey from the European Paediatric Dialysis Working Group. Pediatr Nephrol. doi:10.1007/s00467-015-3137-z
6. Larkins N, Harris KC, Morishita K, Matsell DG (2014) Chylous pericardial effusion in granulomatosis with polyangiitis. Nephrology (Carlton) 19:367–368
7. Rottem M, Fauci AS, Hallahan CW, Kerr GS, Lebovics R, Leavitt RY, Hoffman GS (1993) Wegener granulomatosis in children and adolescents: clinical presentation and outcome. J Pediatr 122:26–31
8. Hoffman GS, Kerr GS, Leavitt RY, Hallahan CW, Lebovics RS, Travis WD, Rottem M, Fauci AS (1992) Wegener granulomatosis: an analysis of 158 patients. Ann Intern Med 116:488–498
9. Forstot JZ, Overlie PA, Neufeld GK, Harmon CE, Forstot SL (1980) Cardiac complications of Wegener granulomatosis: a case report of complete heart block and review of the literature. Semin Arthritis Rheum 10:148–154
10. Litherland J, Lupton EW, Ackrill PA, Venning M, Sambrook P (1994) Computed tomographic peritoneography: CT manifestations in the investigation of leaks and abnormal collections in patients on CAPD. Nephrol Dial Transplant 9:1449–1452
11. Prischl FC, Muhr T, Seiringer EM, Funk S, Kronabethleitner G, Wallner M, Artmann W, Kramar R (2002) Magnetic resonance imaging of the peritoneal cavity among peritoneal dialysis patients, using the dialysate as "contrast medium". J Am Soc Nephrol 13: 197–203
12. Juergensen PH, Rizvi H, Caride VJ, Kliger AS, Finkelstein FO (1999) Value of scintigraphy in chronic peritoneal dialysis patients. Kidney Int 55:1111–1119
13. Macia M, Gallego E, Garcia-Cobaleda I, Chahin J, Garcia J (1995) Methylene blue as a cause of chemical peritonitis in a patient on peritoneal dialysis. Clin Nephrol 43:136–137
14. Chavannes M, Sharma A, Singh R, Reid R, Filler G (2014) Diagnosis by peritoneal scintigraphy of peritoneal dialysis-associated hydrothorax in an infant. Perit Dial Int 34:140–143
15. Tang S, Chui WH, Tang AW, Li FK, Chau WS, Ho YW, Chan TM, Lai KN (2003) Video-assisted thoracoscopic talc pleurodesis is effective for maintenance of peritoneal dialysis in acute hydrothorax complicating peritoneal dialysis. Nephrol Dial Transplant 18:804–808