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Case Report
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Double-Pokemon-Ball Sign in Encapsulating Peritoneal Sclerosis Responsive to Far-Infrared Therapy

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Abstract

Encapsulating Peritoneal Sclerosis (EPS) is a rare but serious complication of peritoneal dialysis. Medical treatment for EPS includes a combination of anti-fibrotic and anti-inflammatory agents. Despite optimal treatment, EPS is still associated with a high risk of morbidity and mortality. We present a case of EPS who was successfully treated with tamoxifen and far-infrared therapy. The abdominal computed tomography scan mimicked the "double-Pokemon-ball sign".

Keywords: Double-Pokemon-ball sign; Encapsulating Peritoneal sclerosis; End-stage renal disease; Peritoneal dialysis; Far-infrared therapy

Background

Gastrointestinal symptoms are common in patients undergoing Peritoneal Dialysis (PD). The differential diagnosis includes peritonitis, Encapsulating Peritoneal Sclerosis (EPS), and all causes of abdominal pain. Peritoneal fluid analysis is essential for definitive diagnosis and imaging studies may be sometimes needed. We report a case of EPS whose abdominal Computed Tomography (CT) scan mimicked the "double-Pokemon-ball sign".

Case Presentation

A 61-year-old man with a history of hypertension, severe mitral regurgitation, type 2 diabetes mellitus, and end-stage renal disease undergoing PD for 9 years was admitted to the hospital with a 2-week history of poor appetite and abdominal fullness on May 19th, 2015. His Tenckhoff catheter was removed due to refractory peritonitis and tunnel abscess on January 20th, 2015, resulting in a transition to hemodialysis. Upon admission, the patient was afebrile (35.2°C) with a heart rate of 67 beats per minute, respiratory rate of 18 times per minute, and blood pressure of 166/76 mmHg. Physical examination showed a distended abdomen and hypoactive bowel sounds. In the peripheral blood, the hemoglobin concentration was 10.0 g/dl, the white blood cell count was 9600 cells/mm^3 with a neutrophil percentage of 69, and the platelet count was 241000/mm^3. The C-reactive protein concentration was 11.0 mg/dl (reference concentration <0.5 mg/dl). The abdominal CT scan revealed a 15 cm × 8 cm × 24 cm locule of fluid accumulation in the right subphrenic space. There was another 9.8 cm × 2 cm × 8 cm locule of fluid accumulation in the left subphrenic space near the surface of the spleen. The coronal view of the bilateral loculated masses mimicked the "double-Pokemon-ball sign" (Figure 1). The axial contrast-enhanced CT image revealed segmental small bowel wall thickening and dilatation (Figure 2A). Based on the patient's history of long-term PD, a tentative diagnosis of EPS was made. CT-guided pigtail catheter drainage of the right-sided subphrenic fluid was performed. 20 ml of bloody fluid was sent for cytologic and microbiologic analysis, and both of the results were negative. The white blood cell count of peritoneal fluid was 16400/mm^3, with a neutrophil percentage of 99. The red blood cell count of peritoneal fluid was 4000/mm^3. His clinical condition and bowel movement improved after the successful drainage and 2-week combination treatment of prednisolone, tamoxifen, and far-infrared therapy. Because of peptic ulcer disease, the patient could only tolerate 10 mg prednisolone per day for 2 weeks and the dosage was rapidly tapered to only 2.5 mg per day as maintenance dose. The follow-up abdominal CT scan in 2017 showed remarkable resolution of ascites (Figure 2B).

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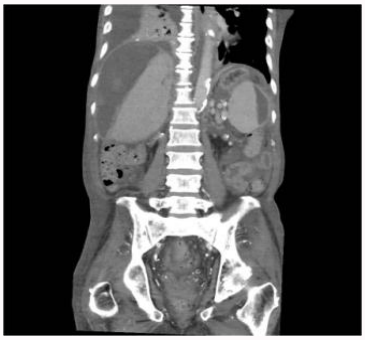

Figure 1: Coronal contrast-enhanced CT image revealed localized fluid accumulation causing splenic and hepatic surface indentation, which mimicked the "double-Pokemon-ball sign".

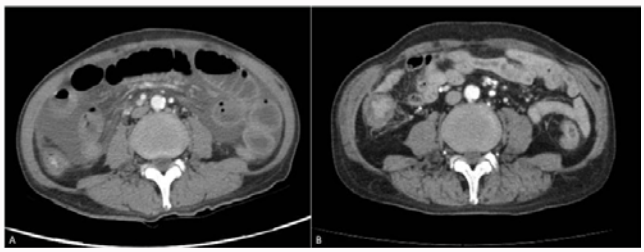

Figure 2A: Axial contrast-enhanced CT image revealed segmental small bowel wall thickening and dilatation.
Figure 2B: Complete resolution of bowel wall thickening and ascites after prednisolone, tamoxifen and far-infrared treatment.

Discussion

EPS is a rare but serious complication of PD, which is characterized by peritoneal inflammation and fibrosis. The incidence of EPS varied widely in different studies and countries and the pathophysiology of EPS is largely unknown. Long PD duration, recurrent peritonitis, and high dialysate glucose concentration have been identified as risk factors for EPS. The common CT findings of EPS include peritoneal thickening/calcification and enhancement, adhesion of bowel loops, bowel wall thickening, signs of bowel obstruction, and loculated fluid accumulation with septum. To the best of our knowledge, this is the first EPS case whose abdominal CT scan mimicked the "double-Pokemon-ball sign".

A combination of tamoxifen and corticosteroid has become the standard treatment for EPS, which significantly improved the patient's outcome. There were previous studies reporting far-infrared therapy in treating EPS, which may be effective because of its anti-inflammatory and endothelium-protective effects [1-3]. Although the suggested dose of prednisolone as standard treatment for EPS may be as high as 0.5 mg/kg/day, the patient was successfully treated with a very low dose of corticosteroids in combination with far-infrared therapy. Further prospective studies are needed to determine the effectiveness of far-infrared therapy for EPS. Surgical intervention such as enterolysis and stripping of the encapsulating membrane is usually reserved for patients with severe obstructive symptoms [4].

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