

Abdominal Cocoon, a Rare Complication of Continuous Ambulatory Dialysis Catheter

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Abstract

Abdominal cocoon, also referred to as sclerosing encapsulating peritonitis, is a relatively rare cause of intestinal obstruction. Pre-operative diagnosis depends on a high index of suspicion. Physicians should be aware of the condition to avoid "surprise" upon laparotomy and to manage cases in a proper way. CT scan is the most helpful tool of investigation. Treatment consists of excision of the accessory peritoneal sac with lysis of the inter-loop adhesions. Bowel resection is unnecessary unless a non-viable segment is found.

INTRODUCTION

Abdominal cocoon, also referred to as sclerosing encapsulating peritonitis, is a relatively rare cause of intestinal obstruction characterized by fibrotic encapsulation of the bowel.

We report a patient with partial intestinal obstruction and abdominal cocoon with an ischemic segment of small bowel diagnosed preoperatively by CT scan. We review the literature and discuss the clinical presentation, investigations and the management of the disease.

CASE PRESENTATION

An 85-year-old man presented to the emergency department at King Fahad Medical City complaining of vomiting and central abdominal pain for three days. He had a history of recurrent attacks of pain for one year which usually resolves spontaneously. He was known to have diabetes mellitus, hypertension, COPD and chronic end-stage renal failure. He was on continuous ambulatory peritoneal dialysis for two years (figure 1) but peritoneal dialysis was substituted last year with haemodialysis due to repeated infections of the peritoneal catheter.

Figure 1

Figure 1: Plain abdominal x-ray of the patient taken 2 years before the last admission showing the CAPD catheter in the pelvis (arrow).



He was on haemodialysis twice weekly for a year. On examination, he was febrile and dehydrated, with a temperature of 38.8°C, a pulse rate of 109 beats/min. and a blood pressure of 115/60 mmHg.

Abdominal examination revealed a tender central abdominal mass extending from the epigastric region to below the umbilicus. The remaining abdomen was soft and not tender. Bowel sounds were absent. Digital rectal examination was unremarkable. Examination of the vertebrae and lower limbs showed scoliosis of thoracolumbar vertebral column associated with severe flexion deformity of both knees and hip joints.

The pre-operative investigations revealed: WBC 9.8×10^3 , neutrophils 79%, Hb 10.7g/dl, platelets 102×10^3 , high creatinine (399) due to ESRD, urea 6.4 mmol/L, sodium 130, potassium 3.6 and markedly elevated serum lactate (5 mmol/L).

The supine abdominal x-ray showed mildly dilated small bowel in the upper abdomen (figure 2).

Figure 2

Figure 2: Plain abdominal X-ray showing dilated loops of bowel at upper abdomen associated with a soft-tissue mass shadow occupying the centre of the abdomen. (Observe the scoliosis of the lumbar spine and osteoarthritis changes of both hips).



Pre operative echocardiogram showed left ventricular dilatation with an ejection fraction of 25% and a dilated left atrium with mild mitral & tricuspid regurgitation and aortic degenerative changes.

The CT scan showed concentration of the small-bowel loops to the center of the abdomen, encased by a thick soft-tissue density associated with mural thickening and air within the bowel wall, suggestive of bowel ischemia (figures 3&4).

Figure 3

Figure 3: CT showing clusters of dilated small-bowel loops enclosed within a thick membrane (arrow).



Figure 4

Figure 4: Mural thickening and air within bowel wall as demonstrated on CT.



Diagnosis of abdominal cocoon was made on the clinical background (history of continuous ambulatory peritoneal dialysis, subacute bowel obstruction, history of similar episodes that resolved spontaneously, and the presence of the abdominal mass), supported by the CT scan findings.

After initial fluid resuscitation and obtaining high-risk consent, the patient underwent an urgent exploratory laparotomy.

Peroperatively, the entire small bowel with the exception of 60cm of proximal jejunum and distal ileum was found to be encased in a cocoon-like fibrous membrane (figures 5&6).

Figure 5

Figure 5: Intra-operative photograph showing the entire small bowel with the exception of 60cm of proximal jejunum and distal ileum encased in a cocoon-like fibrous membrane



Figure 6

Figure 6: Intra-operative photograph showing dilated proximal and distal small bowel with inflammatory exudates over the membrane.



The membrane was peeled off and the small-bowel loop adhesions were freed by blunt dissection. At the centre of the cocoon a segment of small bowel about 70cm in length was found to be ischemic. The ischemic segment was resected with side-to-side anastomosis.

The patient had a stormy postoperative period. He was kept in the ICU on positive pressure ventilation and inotropic support for few days. His bowel function returned on the fifth postoperative day, nasogastric feeding was commenced

and he was gradually weaned of the inotropes. An attempt of extubation failed twice due to desaturation. Unfortunately, he developed severe pneumonia complicated with gram-negative septicemia and septic shock. His post-operative echocardiogram was suspicious of infective endocarditis. In spite of appropriate antibiotic therapy the patient continued to deteriorate and died on the 14th post-operative day.

The histology of the excised sac showed fibrocollagenous tissue with inflammatory reaction. The resected bowel segment showed evidences of ischemia.

DISCUSSION

Abdominal cocoon or sclerosing encapsulating peritonitis is a condition characterized by total or partial encasement of the small bowel by a fibrocollagenous cocoon-like sac (1). The abdominal cocoon was first described by Owtschinnikow in 1907 as “peritonitis chronica fibrosa incapsulata” (2). In 1978, Foo et al. reported 10 cases in whom they could not find any cause for this peculiar type of peritonitis producing intestinal obstruction and they, for the first time, grouped this condition as a new clinical entity “the abdominal cocoon” (1,2,3). Deeb et al., in 1998, described the disease as sclerosing encapsulating peritonitis (4). Since the time this disease was first described, approximately 35 cases have been reported till the year 2007 (5).

Many cases have been reported in the past few years due to the increased awareness of this condition, which appears to be more common than it was previously thought (5, 6, 7). The condition has been classified as primary and secondary based on whether it is idiopathic or has a definite cause.

Primary or idiopathic cocoon occurs in young girls, especially those from tropical and subtropical areas, presenting with small-bowel obstruction and a palpable abdominal mass without any obvious cause. Because of the peculiar age and sex distribution of the disease, it was postulated that the condition is due to retrograde menstruation with subclinical, viral peritonitis resulting in the development of an encapsulating membrane on the intestine (8).

Secondary abdominal cocoon may occur as a serious complication of CAPD (5). The prevalence of abdominal cocoon in patients undergoing CAPD ranges from 0.5% to 2.8% (9, 10). Holland reported sclerosing peritonitis in patients on chronic ambulatory peritoneal dialysis. Dialysate solutions and bacterial peritonitis have been reported as etiological factors. But neither of these hypotheses has been

proven (11).

Some drugs, especially the betaadrenergic blocker ‘practolol’ have been suggested as a possible cause of secondary abdominal cocoon, because it may lead to enhanced collagen production and subsequent fibrosis (12, 13, 14). Abdominal cocoon has also been described in association with sarcoidosis, systemic lupus erythematosus, indwelling abdominal catheters (specifically Le-Veen shunts), orthotopic liver transplantation, and tuberculous pelvic inflammatory disease (2, 4, 15).

The clinical diagnosis of abdominal cocoon requires a high index of suspicion because of the non-specific clinical picture, non-contributory imaging findings (16) and the fact that there are only few reports in the literature on its radiologic imaging findings (17). Clinicians must rigorously pursue a preoperative diagnosis, as it may prevent a “surprise” upon laparotomy and result in proper management (18).

In the cases of abdominal cocoon described in the literature to date, the diagnosis was made either during surgery for unrelated reasons (in asymptomatic patients) or at exploratory laparotomy (in patients who presented with bowel obstruction (16).

The four main clinical features that help identify abdominal cocoon preoperatively as suggested by Yip include intestinal obstruction presenting with abdominal pain and vomiting, but rarely all four cardinal symptoms (intestinal obstruction occurring in a relatively young girl, without an obvious cause, a history of similar episodes that resolved spontaneously, and the presence of a non-tender soft mass on abdominal palpation) are found (19).

Features of recurrent acute or chronic small-bowel obstruction result from kinking and/or compression of the intestines within the constricting cocoon (1, 20, 21). The abdominal mass may result from an encapsulated cluster of dilated small-bowel loops. Progression of bowel obstruction to bowel ischemia and gangrene may result from severe kinking or compression.

The characteristic radiological findings of sclerosing encapsulating peritonitis have been sparsely described (22).

Plain x-rays may show dilated loops of small bowel at the centre of the abdomen. Abdominal ultrasound may show clumping of bowel loops with the bowel surrounded by a thick rim of hypo-echoic tissue. Tethering of the bowel

posteriorly or the presence of a membrane anterior to the small bowel may be seen (23).

The classic barium study findings are a serpentine or concertina-like configuration of dilated small-bowel loops in a fixed U-shaped cluster. Some authors have described a cauliflower-like appearance on barium study (24, 25).

Compared with other imaging techniques, CT gives a more complete picture of this entity as well as of any associated complications, and may also help to exclude other causes of intestinal obstruction (26).

The typical finding of abdominal cocoon on CT is a concentration of the whole small-bowel to the centre of the abdomen encased by a soft-tissue-density mantle (27). Other CT features of abdominal cocoon include signs of obstruction, agglutination and fixation of intestinal loops, mural thickening, ascites and localized fluid collections, peritoneal thickening and enhancement, peritoneal or mural calcifications, and reactive adenopathy (28). Peritoneal encapsulation is a separate entity, which may be confused with abdominal cocoon. It may have a similar appearance to abdominal cocoon on radiologic studies. Peritoneal encapsulation is a congenital condition in which all or part of the small bowel is encased by an accessory peritoneal membrane. Usually, this is asymptomatic, but has been reported to cause bowel obstruction in a few cases. Pathologically, the encasing membrane is normal peritoneum rather than the thick fibrous-collagenous tissue seen in abdominal cocoon (31, 32) which probably derives from plasma exudation from the peritoneal microvasculature (31).

The typical finding at surgery is a conglomeration of small-bowel loops encased in a dense white membrane (32, 33). Treatment consists of excision of the accessory peritoneal sac with lysis of the inter-loop adhesions. Bowel resection is unnecessary (32, 3), unless a non-viable segment is found (33, 34).

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