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## CASE REPORT

# Abdominal Cocoon Syndrome

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## ABSTRACT

***The abdominal cocoon syndrome was first described as a rare condition where part of or the whole small bowel is encased within a fibrous membrane. This report is of a 25-year-old Chinese woman who presented with increasing abdominal distension and jaundice, and was thought to have liver pathology. However, at ultrasound and computed tomography of the abdomen and pelvis, a multiloculated huge cystic structure was seen, extending from the pelvis to the liver with a loop of small bowel encased within. Postoperatively, the diagnosis of an abdominal cocoon was made and a haemorrhagic aetiology was suggested.***

***Key Words:*** *Abdomen, Computed tomography, Fibrous membrane, Peritonitis*

## CASE REPORT

A 25-year-old Chinese woman was referred from a private hospital with a 3-month history of worsening symptoms of ascites of unknown aetiology. A peritoneal tap was done in the private hospital and yielded blood stained ascitic fluid but no malignant cells. Two weeks prior to admission to the University of Malaya Medical Center, the patient developed worsening shortness of breath and became anaemic with a haemoglobin level of 75.8 g/L (normal range, 120 to 150 g/L). Biochemical investigations revealed normal liver function except for a bilirubin level of 31 μmol/L (normal range, 5 to 21 μmol/L) and hepatitis screen was negative. Physical examination revealed a cachexic jaundiced female with gross abdominal distension. The internal organs were not palpable due to gross ascites.

Ultrasound showed the whole abdomen and pelvis to be occupied with numerous multiseptated small cysts. There was a relatively echogenic mass seen in the left adnexa, not separate from the cystic mass. Contrast enhanced computed tomography (CT) scan of the chest, abdomen, and pelvis revealed a large cystic mass arising from the right adnexa and extending to the level of the upper abdomen, between the liver and the right diaphragm (Figure 1). A loop of small bowel was trapped within the cystic mass (Figure 2). The septae seen on ultrasound were not visible. The portal vein was thrombosed with cavernous transformation (Figure 3). The spleen was enlarged to 30 cm. Multiple varices and spleno-renal shunts were seen, in keeping with portal hypertension. The liver was otherwise normal with no dilated ducts. No pleural effusion or ascites was seen. A diagnosis of a large ovarian mass was made. The patient was given 2 units of blood to increase her haemoglobin levels and to stabilise her general condition prior

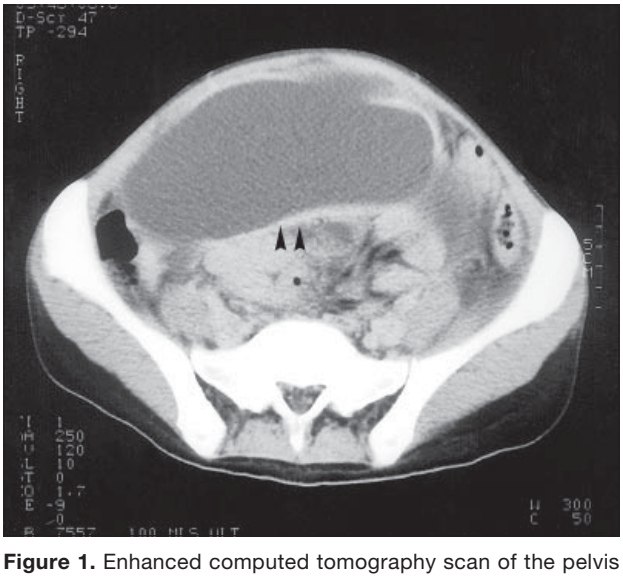


**Figure 1.** Enhanced computed tomography scan of the pelvis showing a cystic mass that seems to be arising from the pelvis.

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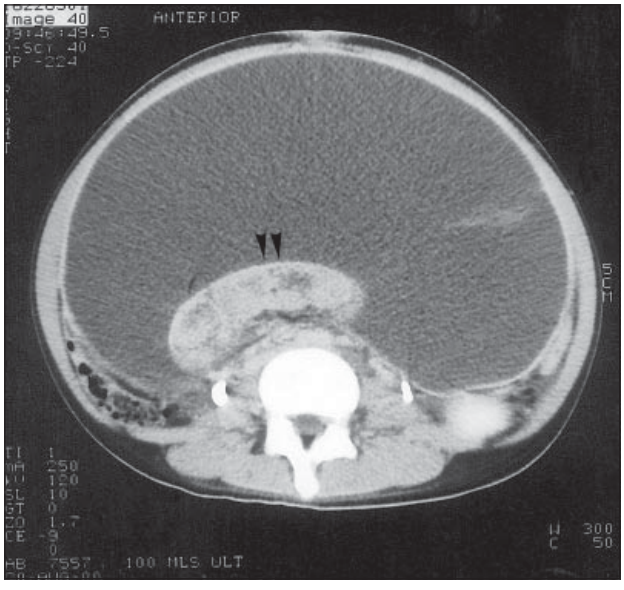


**Figure 2.** Enhanced computed tomography scan showing a loop of bowel encased within a cystic mass.

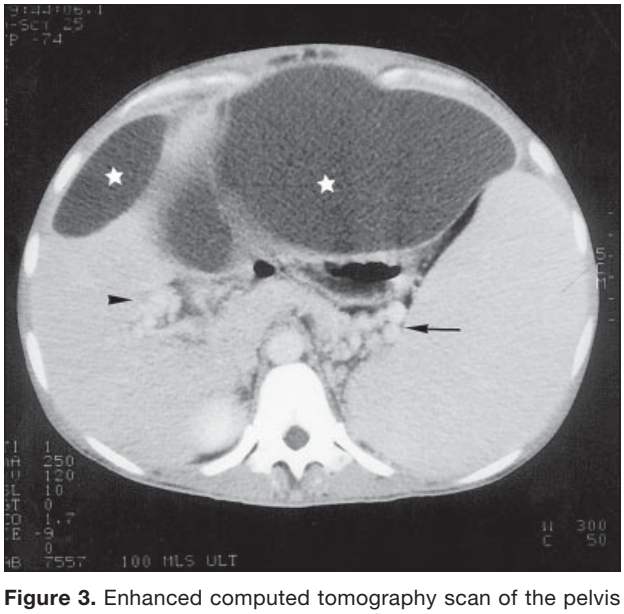


**Figure 3.** Enhanced computed tomography scan of the pelvis showing loculated collections of fluid (★), cavernous transformation of the portal vein (←), and splenic varices (➤).

to operation. At operation, oesophageal varices were noted. Dense adhesions between the liver and the anterior abdominal wall, as well as a large spleen were seen. Seven litres of blood-stained fluid were removed from the peritoneal cavity.

At operation, a large 'cocoon' containing loops of small bowel was found. The membrane of the cocoon was thickened. Locules were broken and a drain inserted. Histological examination of the peritoneal biopsy showed fibrin and necrotic tissue with acute inflammatory cells as well as haemorrhagic tissue. No granuloma or malignancy was seen and no organisms were cultured. Features were consistent with recent and organising haemorrhage. The ascites cleared and the patient was discharged with a haemoglobin level of 108.5 g/L. The postoperative period was uneventful and follow-up 1 month later showed no recurrence of the ascites or jaundice.

## DISCUSSION

Abdominal cocoon is a rare condition that refers to total or partial encapsulation of the small bowel by a fibrocollagenous membrane or cocoon with local inflammatory infiltrate leading to acute or chronic bowel obstruction.[1,2] The condition has been described with various names including 'peritonitis chronica fibrosa incapsulata' by Owtschinnikow in 1907[3] and sclerosing encapsulating peritonitis by Deeb et al in 1998.[2]

The condition has been classified as primary and secondary based on whether it is idiopathic or has a definite cause.[2] The aetiology of the primary form is still uncertain with various hypotheses proposed, although it is probably caused by a subclinical peritonitis leading to the formation of a cocoon.[1,4,5] Foo et al detected the condition in 10 young girls with symptoms of bowel obstruction 2 years after menarche and postulated that a chemical peritonitis was caused by retrograde menstruation, leading to the formation of a cocoon.[4]

Secondary causes include the placement of Le Veen shunt for refractory ascites,[5] continuous ambulatory peritoneal dialysis,[6] systemic lupus erythematosis, and the use of povidone iodine for abdominal wash-out,[7] as well as the β-adrenergic blocker, practolol.[8] Practolol has been withdrawn from use because it was noted to cause the formation of a peritoneal membrane.

Clinically, most patients with abdominal cocoon syndrome present with features of recurrent acute or chronic small bowel obstruction secondary to kinking and/or compression of the intestines within the constricting cocoon.[1,4,5] An abdominal mass may also be present due to an encapsulated cluster of dilated small bowel loops. In this patient, the main symptoms were increasing abdominal distension, jaundice, and features suggestive of liver pathology.

The preoperative barium follow-through study showed characteristic findings of serpentine configuration of dilated small bowel within the cocoon.[1] Using CT, a preoperative diagnosis of abdominal cocoon is likely to

be more easily made. The imaging features are, however, not pathognomic. It has been observed that CT findings of a membrane enveloping loops of small bowel were seen in some paraduodenal hernias, abdominal cocoon, and in peritoneal encapsulation. However, clinical and pathological features of these entities are different.[9] The final diagnosis of abdominal cocoon is usually made based on intraoperative and histopathology findings,[10] with a significant number presenting for emergency treatment without any imaging being performed. In all the reported patients, portions of the small bowel were encased within a fibrous cocoon and, in 1 patient, it was reported to extend and fuse with the parietal peritoneum.[4] Differential diagnosis included peritoneal encapsulation, which was described as a developmental anomaly where the whole of the small bowel is encased in a thin accessory membrane. The clinical symptoms of this condition differ from those of the abdominal cocoon syndrome, in that the patients are mostly asymptomatic and the findings are incidental and late in life.[5] The management of abdominal cocoon is usually removal of the membrane, after which recovery is usually complete, as for this patient.

In conclusion, this report is of a patient with an abdominal cocoon, and is intended to raise awareness of this diagnosis and its imaging characteristics. This should enable earlier preoperative diagnosis and prevent unnecessary bowel resection.

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