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Abdominal Cocoon Syndrome: A Case Report

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Abstract
Abdominal cocoon syndrome (ACS) is defined as idiopathic partial or total encapsulation of the bowel and other abdominal organ within a fibrocollageneous membrane and considered as a rare cause of small bowel obstruction (1). Also known as primary sclerosing encapsulating peritonitis (SEP). The presence of fibrous membranes that encompasses the intestines forming a sort of butterfly cocoon. Patients often present with symptoms of partial intestinal obstruction, which is challenging to diagnose before laparotomy. Of all the available investigations, the contrast-enhanced computed tomography of the abdomen is the most sensitive, showing a sac like fibrous membrane covering the bowel loops along with the fluid collection. Definitive treatment includes excision and adhesiolysis.

Key words: Abdominal cocoon syndrome, intestinal obstruction, sclerosing encapsulating peritonitis.

Introduction

Abdominal cocoon, or "primary sclerosing encapsulating peritonitis", is a rare disease of unknown aetiology, reported in tropical countries, in which the intestine is covered by membranes that cause intestinal obstruction. Peritoneal encapsulation (PE) is a congenital abnormality, in which part or all of small intestine is covered with a thin peritoneal membrane and without intestinal adhesions and rarely characterises by intestinal obstructions (2,3). Sclerosing encapsulated peritonitis (SEP) are two types, Idiopathic and secondary due to tuberculosis, ventriculoperitoneal shunt, povidone peritoneal lavage etc. (2,4). Sclerosing encapsulated peritonitis (SEP), first described in 1907 by Owatschinnikow (5) as peritonitis chonica fibrosa incapsulata; the covering membrane is a greyish-white, thick and fibrous cocoon like membrane (6,7). It was termed 'abdominal cocoon' by Foo in 1978 (8,9,10). It is challenging to diagnose abdominal cocoon syndrome preoperatively, and therefore it is frequently misdiagnosed (11,12,13).
The underlying aetiology is unknown and primarily seen in adolescent females from tropical and subtropical areas, although cases have been reported in premenopausal women, children and males (6,7,8). Male female ratio is 1:14. However we present a male case here.

Case Report:

A 65-year old male patient presented with abdominal pain, vomiting for two days at accident and emergency department of RIPAS Hospital at Bandar Seri Begawan, Brunei. No history of fever or chills, no obstructive urinary symptoms. On his examination there was tenderness at centre and supra pubic area of abdomen, no guarding and bowel sounds was positive. Pain was crampy and gripping in nature. He has persistent hiccup, reduced appetite and didn't take any food for two days. No history of blood/black stool. Previous endoscopy showed gastritis and mild esophagitis and colonoscopy showed benign transverse colonic polyp one year back. The patient was advised for x-ray abdomen. On x-ray there was no sign of perforation and bowel loops filled with faecal matter. Ultrasound of abdomen showed no mass. The patient was admitted and advised for CT scan of abdomen. CT scan findings showed clusters of small bowel loops at mid

abdomen which were partially surrounded by a thin enhancing membrane forming a sac- suggestive of abdominal cocoon syndrome (Fig 1,2 &3). Laparotomy was done with adhesiolysis and excision of cocoon. After surgery patient's condition improved and was discharged home. Histopathological report showed fibrous tissue.

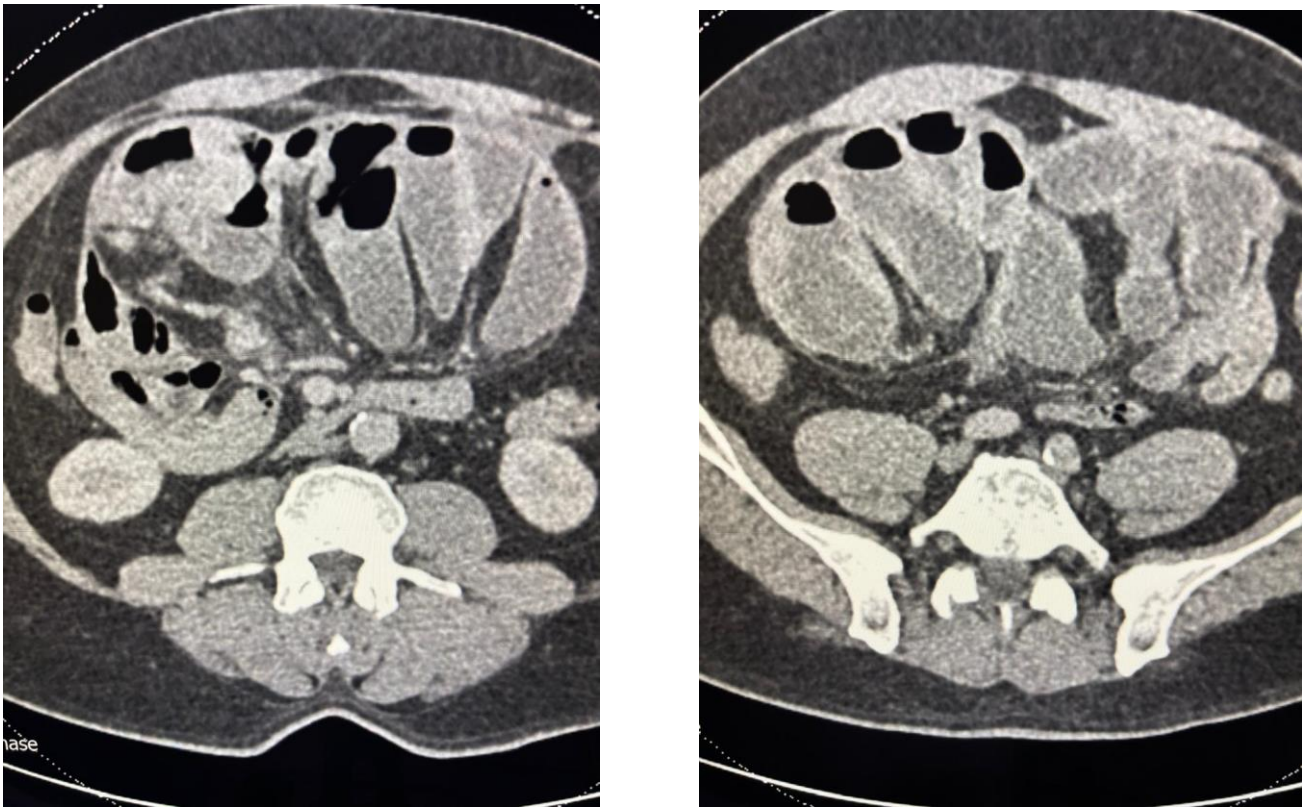

Figure 1: Axial CT scan of abdomen

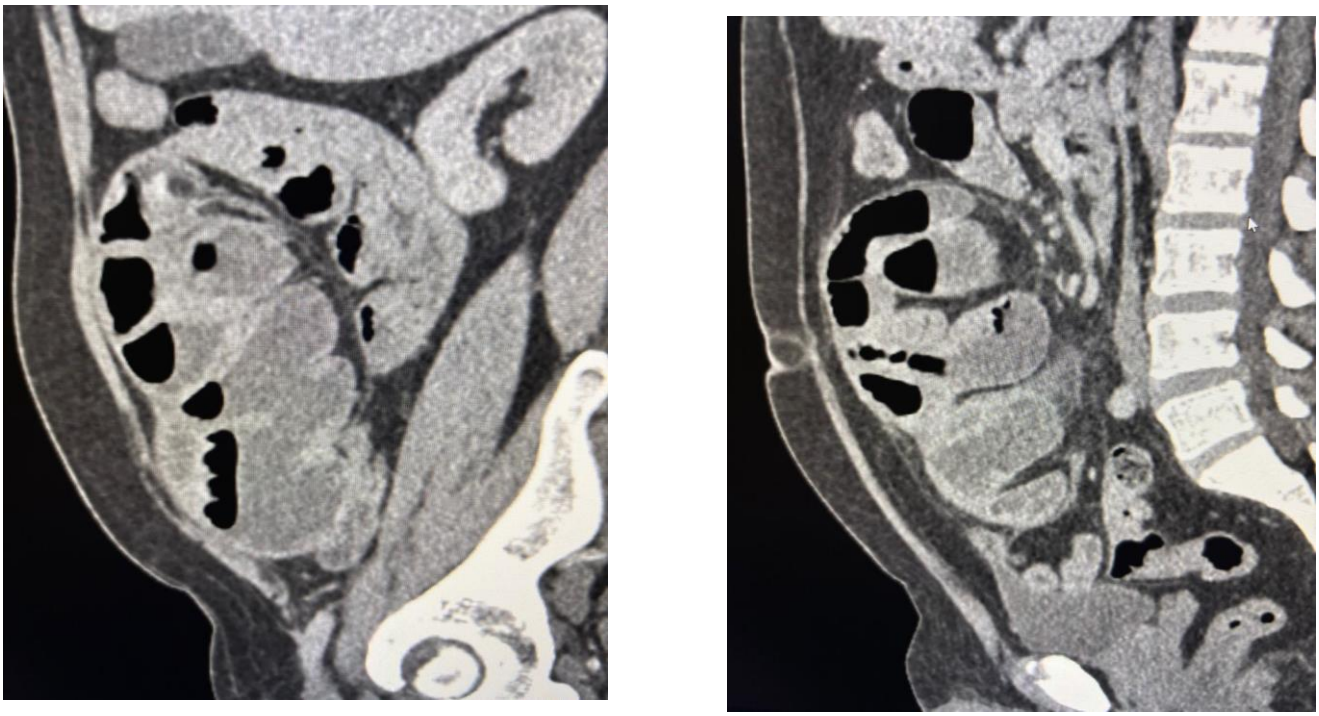

Figure 2: Sagittal CT scan of Abdomen

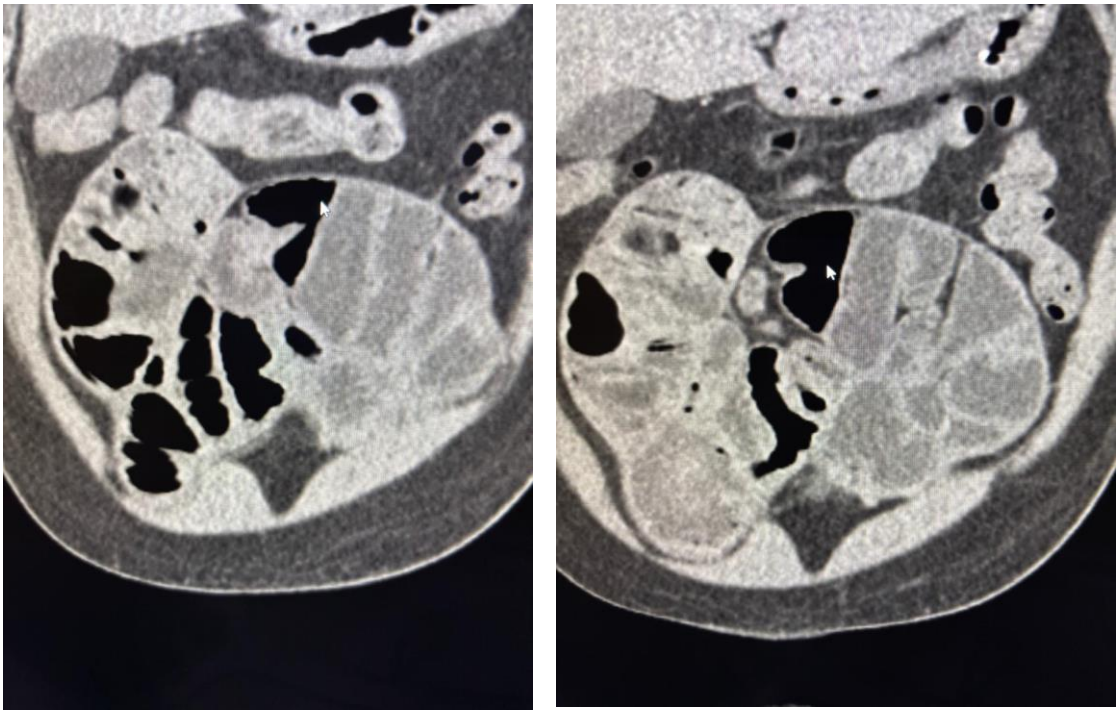

Figure 3: Coronal CT scan of Abdomen

Discussion

Sclerosing encapsulating peritonitis (SEP) is a rare cause of abdominal pain and intestinal obstruction. Primary SEP or idiopathic SEP – commonly referred to as abdominal cocoon, has no specific aetiology. Some theories were postulated to describe aetiology, retrograde infection, embryological abnormality, mesenteric hyperplasia, mesenteric vessels malformation, in addition, microbial cell-mediated tissue damage is seen in some cases (14).

Secondary SEP has multiple aetiologies continuous ambulatory peritoneal dialysis, peritonitis, tuberculosis, previous surgery, sarcoidosis, abdominal shunt, lymphoma, SLE etc. PE is also associated with intestinal and colonic malrotation, cryptorchidism, hernia (15). Other rare causes can be idiosyncratic drug reactions, postsurgical sequelae, abdominal trauma, autoimmune disease. Few cases are also associated with hepatitis C, liver transplantation, free abdominal gas etc. These may lead to chronic peritoneal irritation and inflammation, leading to peritoneal fibro neurogenesis

Types of abdominal cocoons

Type-I : Only a part of small intestine
Type II : Entire small intestine
Type III : Entire small bowel and colon or other organs such as liver, stomach, ovaries etc
Type IV : Involves the entire peritoneum (also known as advanced type III)

Histopathology of SEP shows dense fibrocollageneous connective tissue with hyalinization, sparse capillaries network and chronic inflammatory infiltrate, with or without lymphocytic or plasma cell.

A SEP can present with recurrent episodes of abdominal pain, anorexia, nausea, vomiting, weight loss, acute /subacute small bowel obstruction. Some patient has palpable abdominal mass, some are asymptomatic. Abdominal X-ray may be normal or clumps of small bowels are seen. Ultrasound may show aggregated small bowel, ascites rarely covering membrane and adhesions. Barium follow through shows clumped loops towards the centre of abdomen may appear like cauliflower.

A CT scan with contrast is more sensitive. It shows a sac like structure of fibrous membrane covering the bowel loops- 'Cauliflower sign'. A capsule may not appear in CT. MRI is also useful as the membrane is more evident than on CT.

In patient with mild symptoms, conservative management is given with adequate hydration, NG tube decompression, and bowel rest. As medications tamoxifen (for patient with dialysis), immunosuppressants, steroids etc are used. In recurrent and obstructive cases surgical excision, adhesiolysis are performed.

Post-operative complications include infection, small bowel obstruction, short bowel syndrome, perforation, intestinal fistula etc.

Conclusion

Our aim of this case report is to highlight the importance or raise awareness of abdominal cocoon as a possible rare cause of abdominal pain and subacute intestinal obstruction after excluding all other possible common causes. Detailed history and physical examination are helpful especially in case of secondary SEP. Contrast enhanced computed tomography is a valuable tool for preoperative diagnosis. However, in most cases, it is diagnosed intraoperatively and proved with postoperative histopathological findings. Surgical treatment is the main option.

Conflict of interest: None.

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