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Case Report

Abdominal cocoon syndrome: diagnostic challenge in an uncommon entity

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ABSTRACT

Abdominal cocoon syndrome (ACS) also known as sclerosing encapsulating peritonitis (SEP) is a rare condition where thickened fibro-collagenous peritoneal membrane encases parts of the small intestine likely due to recurrent inflammatory processes. Aetiology is unidentified in most of the cases but it is notably associated with patients on chronic peritoneal dialysis for end stage renal disease. Diagnosis is usually intraoperative. However, CECT abdomen is a definitive radiological imaging modality of choice. SEP can be managed both conservatively as well as surgically. Here, we present a case of 48-year-old female patient presenting with features of small bowel obstruction and sepsis and was a known case of chronic liver disease (CLD). She had history of episodes of recurrent small bowel obstruction. She was managed conservatively for the same. She was diagnosed with the help of CECT abdomen and was managed conservatively in view of high-risk surgery due to poor general health and comorbid conditions. She could not survive and succumbed to liver dysfunction and sepsis.

Keywords: Cocoon abdomen, Sclerosing encapsulating peritonitis, Chronic liver disease, Conservative management, Intestinal obstruction

INTRODUCTION

Sclerosing encapsulating peritonitis (SEP) often addressed as cocoon abdomen is an infrequent disease entity characterised by formation of thick fibrotic membrane which partially or completely encapsulates the bowel loops.[1] Owtschinnikow was the first to document this condition in 1907.[2] It can be primary where cause is unknown or secondary due to predisposing factors such as use of beta-blockers, liver cirrhosis, abdominal tuberculosis, and previous abdominal surgery, peritoneal dialysis, ventriculoperitoneal shunts.[1,3] Idiopathic SEP is usually more common in young females, but SEP can occur at any age. Its presentation ranges from asymptomatic patients to non-specific symptoms such as vomiting, abdominal pain and/or subacute bowel obstruction.[3] Radiological investigations done are X ray abdomen erect, ultrasonography, CT scan. Contrast enhanced CT scan abdomen is the most useful diagnostic modality. It is managed conservatively as well as surgically. Some of the cases of SEP may be diagnosed intraoperatively.

CASE REPORT

A 48-year-old postmenopausal female presented to surgical department of our hospital with chief complaints of abdominal pain, distension, vomiting for 5 days and inability to pass stool or flatus for the past 3 days. She was a known case of chronic liver disease (CLD) for 10 years. There was a history of recurrent episode of subacute small bowel obstruction managed conservatively previously. On examination, she was cachexic, hypotensive, tachycardic, and icteric and needed oxygen support. Abdominal exam showed distension in epigastric region with clumped palpable bowel loops, generalised tenderness, sluggish bowel sounds, and mild ascites. Her plain abdominal X-ray film showed dilated bowel loops in epigastric region (Figure 1). Contrast-enhanced CT of the abdomen showed

clustered, dilated small bowel loops encased within a thick fibrous membrane, findings indicative of abdominal cocoon. No frank signs of perforation or ischemia were noted. Mild ascites and changes of liver parenchymal disease were seen. Due to poor general condition and decompensated liver disease, surgery was considered too high-risk. A conservative approach with bowel rest, IV fluids, nasogastric decompression, antibiotics, albumin infusion and whole blood transfusion was started. She required 4 cycles of haemodialysis for worsening renal functioning. Despite intensive supportive care, the patient's condition worsened. She developed worsening hepatic encephalopathy, renal dysfunction, and sepsis. She passed away on day 6 of admission due to multiorgan failure secondary to septic shock on the background of decompensated CLD.

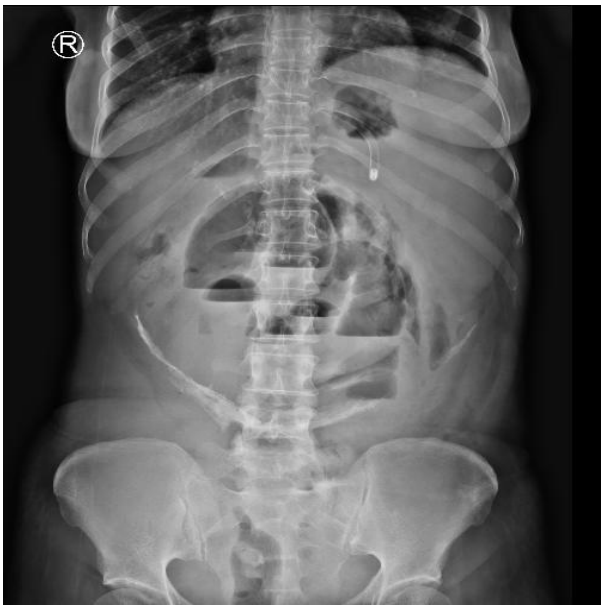

Figure 1: X-ray abdomen film.

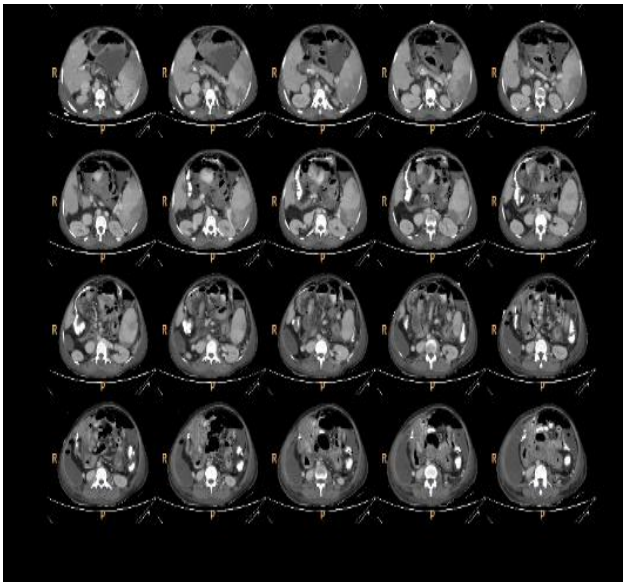

Figure 2: CECT abdomen.

Table 1: Blood investigations.

Investigations	Patient value	Normal range
Hb (gm/dl)	7.6	12-14
Total leucocyte count (U/L)	16300	4000-10000
Platelets count (U/l)	112000	150000-410000
Total protein (gm/dl)	4.7	6-8.3
Albumin (gm/dl)	2.4	3.5-5.5
PT/INR (mins)	23.2/2.3	12-16/1-1.2

DISCUSSION

Sclerosing encapsulating peritonitis (SEP) is a rare acquired abnormality.[1] The condition was initially described by Owtschinnikow et al in 1907, who referred to it as peritonitis chronica fibrosa incapsulata.[2,4] The term 'abdominal cocoon' was later designated by Foo et al, in 1978.[5] According to recent literature, approximately 240 cases have been documented globally.[6]

Abdominal cocoon is grouped into two forms: primary, which has an unidentified origin and is usually seen in young females, and secondary, which occurs due to identifiable risk factors such as beta-blocker use, liver cirrhosis, abdominal tuberculosis, prior abdominal surgeries, peritoneal dialysis, or ventriculoperitoneal shunting.[1,3] In a patient with abdominal tuberculosis, peritoneal involvement typically results from the dissemination of bacilli originating in the mesenteric lymph nodes.[7] It is further classified into three types based on the extent of intestinal involvement; Type I involves partial encapsulation of the small intestine; Type II is characterized by complete encasement of the entire small intestine; and Type III involves not only the encasement of the entire small intestine but also includes other intra-abdominal organs, such as the appendix, cecum, ascending colon, or ovaries.[8]

Our patient was a middle-aged female, diagnosed case of chronic liver disease and presented with typical symptoms of recurrent intestinal obstruction. She was diagnosed using contrast enhanced CT scan of abdomen (Figure 2). Presentation can be acute, subacute or chronic. Most common presentation of this condition is small bowel obstruction and patient presents with abdominal pain, distension, vomiting and obstipation.[9] Very important differential diagnosis for this condition is peritoneal encapsulation which is formation of thin additional peritoneal membrane encases portions of the small intestine. It is usually asymptomatic and diagnosed incidentally, it can sometimes present with symptoms of small bowel obstruction.[10] Plain abdominal radiographs lack specificity, though they may show air-fluid levels or dilated bowel loops suggestive of bowel obstruction. In contrast, computed tomography offers greater diagnostic accuracy, often demonstrating clustered small bowel loops centrally within the peritoneal cavity, surrounded

by a thick encapsulating membrane.[9] Though many cases are diagnosed intraoperatively, CECT abdomen remains gold standard for the diagnosis. Procalcitonin is a useful biomarker for identifying bacterial sepsis in patients presenting with clinical symptoms.[11] Management includes conservative therapy with patient nil per oral, Ryles tube insertion, indwelling urinary catheters, IV fluids, analgesics and antibiotics.[2] In patients with sclerosing encapsulating peritonitis (SEP), nutritional status is a critical consideration. Optimizing nutrition may improve the effectiveness of conservative treatment and reduce the risk of postoperative complications, including infections and anastomotic leaks. Medical management may also include use of steroids.[10,12]

Surgery remains the primary treatment approach for SEP patients who exhibit signs of intestinal ischemia or do not respond to conservative therapy.[2] Surgery involves total removal of the fibrotic membrane along with adhesiolysis to reduce recurrence. SEP is associated with a relatively high mortality rate, reported between 26% and 58%, with malnutrition and sepsis being the leading contributing factors.[13] Surgery was considered too high-risk and was avoided in our case due to poor general condition and decompensated liver disease. Patient succumbed to multiorgan failure secondary to septic shock with decompensated CLD.

CONCLUSION

This case highlights the complexity of managing cocoon abdomen in the context of chronic liver disease. Early diagnosis is crucial, but the presence of comorbidities may limit treatment options. Timely diagnosis and appropriate clinical management are typically associated with positive outcomes. However, the presence of comorbidities such as liver disease may negatively impact prognosis.

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